SUITABILITY OF CHRONIC DISEASE MANAGEMENT IN GENERAL PRACTICE FOR PATIENTS WITH INTELLECTUAL DISABILITIES

MILOU VAN DEN BEMD

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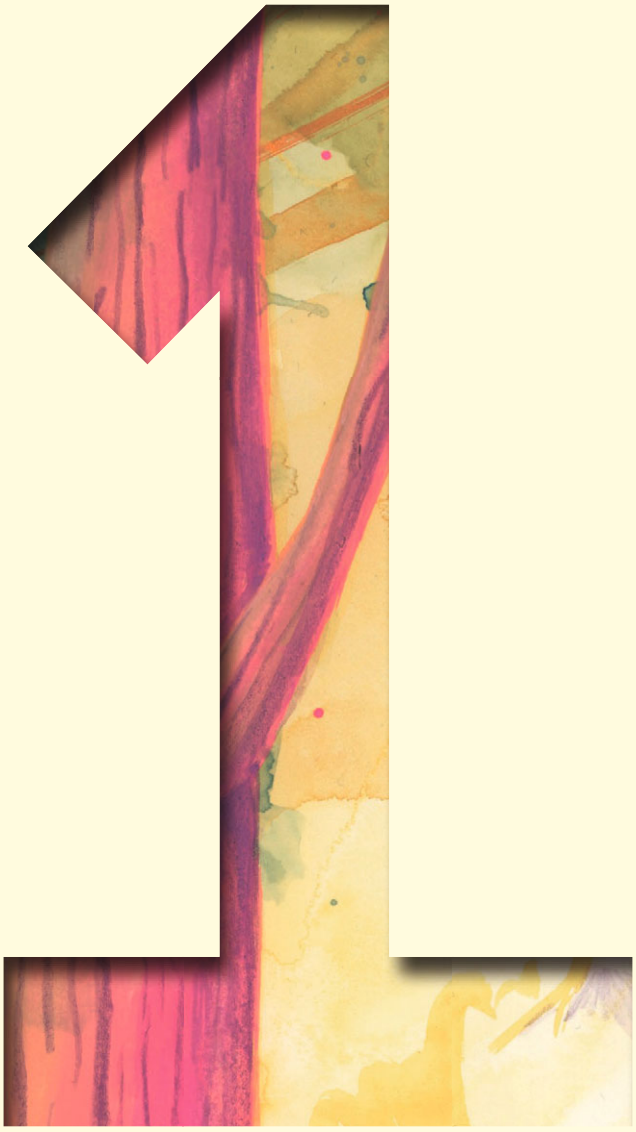
TABLE OF CONTENTS

Chapter 1	General introduction	9
Part I The impact of chronic diseases: prevalence		
Chapter 2	Exploring chronic disease prevalence: A scoping review van den Bemd, M., Cuypers, M., Bischoff, E.W.M.A., Heutmekers, M., Schalk, B.W.M., Leusink, G.L. Published in Journal of Applied Research in Intellectual Disabilities, 2022 doi:10.1111/jar.12957	23
Chapter 3	Chronic diseases and comorbidities: A cross-sectional study in general practice van den Bemd, M., Schalk, B.W.M., Bischoff, E.W.M.A., Cuypers, M., Leusink, G.L. Published in Family Practice, 2022 doi:10.1093/fampra/cmac042	65
Part II Chronic disease management		
Chapter 4	Records of quality indicators: Disease monitoring in residential care Van den Bemd, M., Suichies, V., Leusink, G.L., Bischoff, E.W.M.A., Cuypers, M. Published in British Journal of Learning Disabilities, 2023 doi: 10.1111/bld.12570	91
Chapter 5	Disease monitoring within and outside disease management programmes van den Bemd, M., Cuypers, M., Schalk, B.W.M., Leusink, G.L., Bischoff, E.W.M.A. Published in British Journal of General Practice, 2023 doi: 10.3399/BJGP.2023.0029	115
Part III	Care needs	
Chapter 6	Care needs of chronically ill patients: Patients' and providers' perspectives Van den Bemd, M., Koks-Leensen, M., Cuypers, M., Leusink, G.L., Schalk, B.W.M., Bischoff, E.W.M.A. Revisions submitted	135

Chapter 7	General discussion	163
Chapter 8	Summaries	177
	English summary	179
	Nederlandse samenvatting	183
Chapter 9	Data management statement	189
Chapter 10	Dankwoord	195
Chapter 11	About the author	203
	Curriculum vitae	205
	Portfolio	207
	List of publications	211



GENERAL INTRODUCTION

GENERAL INTRODUCTION

Reducing the impact of chronic diseases in general practice: comprehensive disease management

Globally, chronic diseases are becoming increasingly more prevalent, putting high burdens on healthcare systems and on individuals [1-3]. The highest-impact chronic diseases are cardiovascular disease (i.e., angina pectoris, myocardial infarction, ischaemic heart disease, coronary heart disease, ischaemic or haemorrhagic stroke, or transient ischaemic attack), diabetes mellitus type 1 and 2, and chronic obstructive pulmonary disease (COPD; i.e., chronic bronchitis, bronchiectasis). These chronic diseases are among the most prevalent diseases [1, 4], are among the most common causes of mortality [5], and have some of the highest economic consequences worldwide [2].

Because of these substantial consequences, these chronic diseases require adequate and generally intensive disease management, aimed at preventing complications and comorbidities [6]. They demand regular self-management (i.e., injecting insulin at home), self-care (i.e., maintaining a healthy lifestyle), and regular check-ups and tests. These demands can affect patients' lives as severely as the chronic illness itself [7], since chronic diseases may limit patients' abilities for self-care, recreation, and social participation, resulting in lower quality of life and mental well-being [3, 8, 9].

Most patients in the Netherlands receive chronic disease management in general practice, defined as an 'organised, proactive, multi-component, patient-centred approach to healthcare delivery' for people with a specific chronic disease [6]. It covers multiple interventions across the chronic-disease continuum to prevent comorbidities and complications and ultimately to provide high-quality care to chronically ill patients [6]. General practice aims to guarantee this high quality in at least three ways.

First, the high access to Dutch primary care, and thus general practice, can remove barriers to patients contacting primary healthcare providers in early stages of health problems. As primary care is the first point of access for patients, providing continuous, coordinated, and comprehensive care, general practitioners (GPs) function as gatekeepers to secondary care [10, 11]. In providing chronic disease management, they serve as medical experts on the chronic disease, educators, and lifestyle consultants to support self-management [12].

Second, in the Netherlands, most chronic disease management is delegated to practice nurses, who work under GP supervision [13]. With an extensive range of tasks, including patient education, monitoring disease progression, and signalling complications, they are important in relieving GPs' workload [13]. Their involvement in chronic disease management has also been proved to increase quality of care [14-16].

Third, the quality of chronic disease management is safeguarded by disease management programmes. Since 2010, disease management programmes for diabetes mellitus type 2, COPD, and cardiovascular risk management have been in place [17]. In line with national evidence-based care standards that describe what is considered high-quality care [18], regional healthcare groups have developed and implemented regional or local disease management programmes [19]. A yearly national benchmark report allows healthcare groups to reflect on, and compare, their care provision with national figures, to incentivise them towards high-quality chronic disease management [17].

Within this rather standardised nature of chronic disease management in Dutch general practice, standard modules to some extent can be adapted to patients' care needs, contributing to empowering patients to participate actively in their treatment [17]. Patients are thus included in decision-making processes to ensure treatment adherence and adequate self-management. Decision tools may aid patients to choose treatment options with their healthcare provider, often ranging from lifestyle modifications to more intensive treatments, such as inhalation medication or insulin injections [20, 21].

Chronic disease management for people with intellectual disabilities

People with intellectual disabilities (ID) are characterised by substantial limitations in intellectual functioning and adaptive behaviour as expressed in lower practical skills (e.g., activities of daily living), conceptual skills (e.g., language, money), and social skills (e.g., verbal communication) [22, 23]. ID onset always occurs during the developmental period [24]. Around 187,000 people in the Netherlands – 1.5% of the Dutch population – are estimated to have an ID. Almost half of them receive care in residential care settings, often by GPs employed by the care institution; the other half receive care in regular general practice [25]. With decentralisation of care provision, small-scale residential facilities in the community have increasingly replaced large-scale residential facilities. Therefore, it is likely that an increasing number of people with ID receive care in regular general practice. Furthermore, an additional 1.1 million people (6.4% of the Dutch population) have characteristics of ID, such as low IQ (<85) and problems in adaptive functioning [26, 27] but are not (yet) diagnosed with ID.

The characteristics of ID indicate that this group of people has increased chronic disease risk factors: they have higher rates of unhealthy lifestyles, obesity, and medication use [28-30], often have worse health than people without ID, and are more often hospitalised for avoidable health conditions. Additionally, frailty can occur in them 15 years earlier than in people without ID, and they suffer more often from premature and avoidable mortality [31-36].

Despite these risk factors, it is unclear how often chronic diseases occur in people with ID. Prevalence rates vary highly across studies: for instance, estimates for the prevalence of diabetes mellitus in people with ID range from 0.4% to 25% [37]. In addressing chronic disease prevalence, research also does not often acknowledge the different age and sex distributions between people with and without ID (e.g., males have ID more often than females; the life expectancy of people with ID is lower than that of people without ID, meaning that fewer people reach the age at which people without ID become more at risk for chronic diseases [27, 32]). This may also impact the prevalence of chronic diseases. Accurate information on chronic disease prevalence is essential to facilitate early and timely detection of diseases and to plan the size and allocation of healthcare resources [38].

Additionally, besides limitations in intellectual functioning and adaptive behaviour, other factors complicate suitable care provision. As people with ID often have difficulties understanding and communicating their (symptoms of) disease [39], healthcare providers need additional skills to manage their chronic diseases adequately. Performing physical examinations and adequately transferring information to people with ID is often deemed more complex within the standard short timeframe of consultations in general practice [36, 40]. Standard information regarding medication, disease management, or (the necessity of) screening procedures is often too difficult for them to understand [41]. Existing decision tools, such as choosing inhalators when starting with inhalation medication for patients with COPD, or treatment options for patients with diabetes mellitus [20, 21], are therefore often too complicated for patients with ID. Accessible information, with easy language or visual cues, is not always available [42]. Patients' ability to adequately self-manage their health condition is limited by their high reliance on caregivers or family for support, who often are not trained in providing chronic disease management [43].

Because of these differences from the general population, it is unclear whether disease management programmes are suitable for patients with ID. These programmes, designed to guarantee high-quality chronic disease management [17], are relatively standardised and based on a one-size-fits-all approach, leaving limited room for

healthcare providers to adapt approaches. Additionally, although this standardised nature incentivises healthcare providers towards high-quality care, in less standardised settings, such as residential general practice, chronic disease management may be of lower quality. The lack of incentives in this setting might cause incorrect or missing registrations in patients' medical records. Neither insights into (lack of) recordings of chronic disease management in residential general practice, nor insights into differences between people with and without ID in enrolment and disease monitoring in these care programmes in regular general practice, have yet been provided.

Meeting patients' needs is even more critical when patients have a chronic disease. The difficulties that healthcare providers experience in care provision, as well as the limitations of patients with ID, may then be amplified, because chronic diseases are continuous in nature. Given the differences between people with and people without ID, information on the care needs of people with ID allows healthcare providers to adapt (communication) approaches accordingly. Patients' and healthcare providers' perspectives are essential for appropriate approaches and to prevent both comorbidities and complications [6]. However, these insights are largely lacking, as care needs of patients with ID and their healthcare providers in the context of chronic disease management have not yet been researched.

Aims of this thesis

The characteristics of ID, and concomitant difficulties with comprehending information and limited abilities for self-management, raise the question of the extent to which chronic disease management in general practice is suitable for patients with ID. Inadequate chronic disease management sustains the health disparities between people with and without ID, leaving chronically ill patients with ID at increased risk of complications, avoidable hospital admissions, and premature mortality [31, 35].

This thesis therefore aims to describe the prevalence of chronic diseases, care provision, and care needs of people with ID, with the overall aim of examining the suitability of chronic disease management in general practice for this patient population. More specifically, the research objectives of this thesis are:

(1) To explore the broadness and characteristics of published literature on chronic disease prevalence in people with versus without ID;
(2) To examine chronic disease prevalence and comorbidity patterns by age and sex in people with versus without ID;
(3) To examine the current state of affairs in recordings of chronic disease management in long-term care settings for people with ID;

(4) To examine differences between chronically ill patients with and without ID in enrolment in disease management programmes and disease monitoring;
(5) To explore the needs of patients with ID and of healthcare providers in the context of chronic disease management for patients with ID.

Research setting

Funding was provided by the Dutch Ministry of Health, Welfare, and Sport (grant no. 329574) and the programme for Academic Collaborative Centres for Intellectual Disability of the Netherlands Organisation for Health Research and Development (grant no. 641001100). This research was conducted within the academic collaborative Stronger on your own feet (*Sterker op eigen benen*), a collaboration between six care organisations for people with ID and the research group Intellectual disability & Health of Radboud university medical centre in Nijmegen. The main researcher met several times a year with a co-researcher with ID and an advisory board of professionals working with people with ID (i.e., GP, practice nurse, formal caregiver, ID physician, physician assistant, dietician, and behavioural scientist). They assisted in all phases of research (e.g., study design, formulating interview guides, participant recruitment, interpretation of results, and knowledge dissemination).

Outline of this thesis

Figure 1 provides an overview of this thesis. Firstly, the impact of chronic diseases in terms of prevalence is explored. **Chapter 2** includes a scoping review on chronic disease prevalence in people with versus without ID. In this chapter, prevalence rates of chronic diseases in the literature are described, and differences in prevalence rates across studies are explored. In **Chapter 3**, the prevalence of chronic diseases and comorbidity patterns in people with and without ID is examined. Two studies on disease monitoring then follow. **Chapter 4** explores recordings of quality indicators for chronically ill patients with ID. In **Chapter 5**, enrolment in disease management programmes and disease monitoring in people with and without ID are described. **Chapter 6** includes findings from an explorative study on patients' and healthcare providers' care needs in chronic disease management in general practice. **Chapter 7** provides a general discussion of the main findings as presented in this thesis and recommendations for practice and future research. In **Chapter 8**, all the findings are summarised in both English and Dutch. The data management statement can be found in **Chapter 9**. In **Chapter 10** the acknowledgements can be read. Finally, information about the author can be found in **Chapter 11**.

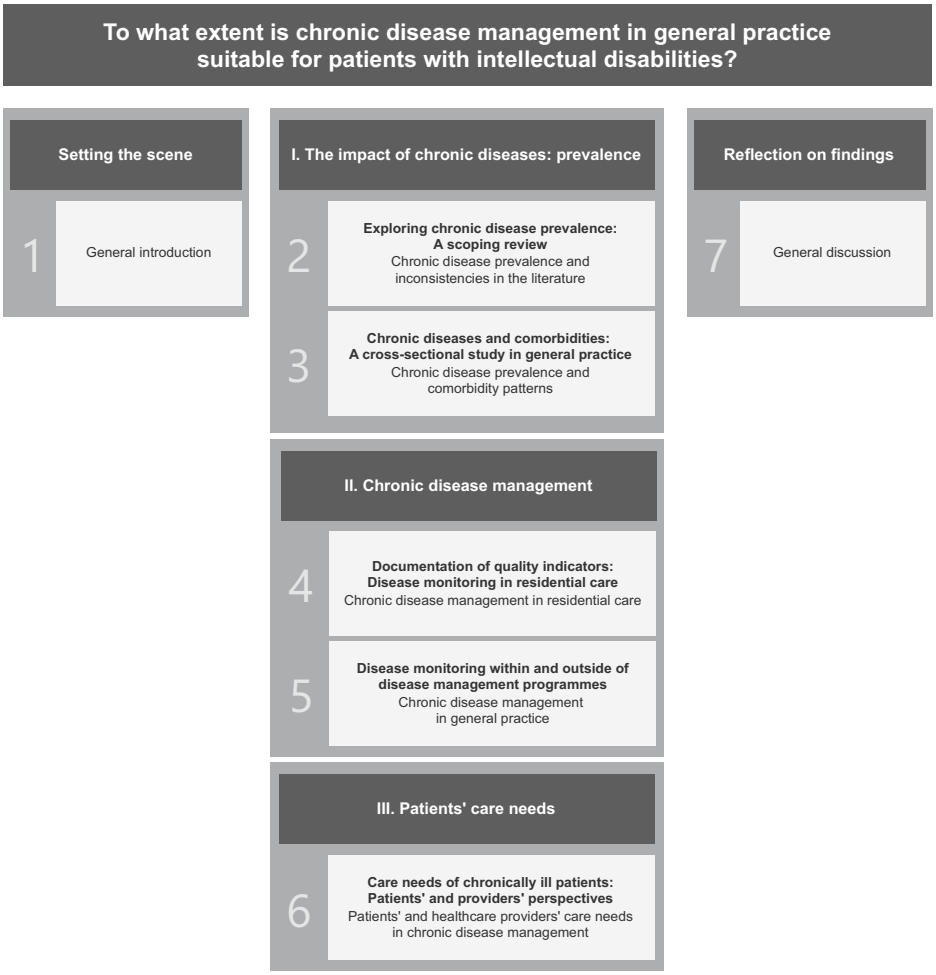


Figure 1. Schematic overview of thesis

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PART I

THE IMPACT OF CHRONIC DISEASES: PREVALENCE

EXPLORING CHRONIC DISEASE PREVALENCE: A SCOPING REVIEW

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ABSTRACT

Introduction: Primary care providers require accurate evidence on chronic disease prevalence in people with intellectual disabilities in order to apply this information into practice. This study aimed to map the broadness of literature on chronic disease prevalence in people with and without intellectual disabilities, and to explore main characteristics of these studies.

Method: A scoping review of peer-reviewed literature was conducted, covering 2000 to February 2020, including literature that discussed chronic disease prevalence in people with and without intellectual disabilities, with similar data collection method for both groups.

Results: Nineteen studies were included. Chronic disease prevalence varied considerably between people with and without intellectual disabilities. Studies differed in their methodologies, country and age groups that were enrolled.

Conclusions: Primary care providers should interpret results on disease prevalence among people with intellectual disabilities in light of the study characteristics. Researchers should always interpret prevalence rates in the context of methodology.

INTRODUCTION

Unambiguous information on chronic disease prevalence in people with intellectual disabilities is largely lacking [1, 2]. Varying and sometimes even conflicting prevalence rates are presented in the literature [2, 3]. Heterogeneity between studies can potentially be reflected in various factors such as sample size, type of data, or methods of identification of intellectual disabilities, making correct understanding and interpretation of chronic disease prevalence in people with intellectual disabilities more complex.

Primary care providers and actors in public health planning require accurate information on chronic disease prevalence to interpret results in terms of chronic diseases being more or less prevalent among people with intellectual disabilities as compared to people without intellectual disabilities [4-7]. Such accurate evidence, that can be applied and translated into practice, is a first necessity in providing optimal healthcare [8]. A better insight into the aspects that relate to the inconsistencies in the literature is therefore necessary to help primary care providers and researchers to better understand and accurately interpret prevalence rates of chronic diseases in people with intellectual disabilities.

Chronic diseases such as ischemic heart disease (IHD), cerebrovascular disease (CVD), diabetes mellitus (DM), and chronic obstructive pulmonary disease (COPD) are among the most common chronic diseases worldwide [9]. They have the highest impact on both the economic level [10, 11] and patients' individual level, such as their quality of life [12-14] and risk of mortality [15, 16]. The impact of chronic diseases can be even higher for people with intellectual disabilities compared to the general population, as they experience limitations in adaptive behaviour and intellectual functioning [17]. As a result, it is more difficult for them to fully comprehend the implications of chronic diseases, and this complicates disease management and results in poorer health outcomes [18].

As chronic diseases are mostly managed in primary care, this setting provides the most complete representation of everyone in the population with and without chronic diseases [19, 20]. Secondary care settings typically report higher prevalence estimates than primary care settings do, as patients in this setting are more likely to have a chronic illness but may be overrepresented by severe cases [19]. It is therefore most relevant to focus on prevalence studies on people with and without intellectual disabilities conducted in primary care settings. Information on the prevalence of diseases such as IHD, CVD, DM, and COPD is also used to plan the size and the allocation of healthcare

resources [21]. Accurate understanding of published prevalence rates is therefore essential. This scoping review therefore aims 1) to map the broadness of published literature on IHD, CVD, DM, and COPD prevalence in people with intellectual disabilities compared to people without intellectual disabilities in primary care settings, and 2) to explore main characteristics of these studies.

METHODS

Study design

This study is a scoping review, a type of review commonly used to map existing literature that "exhibits a large, complex, or heterogeneous nature" [22]. They are particularly useful for describing research findings in more detail by taking different research designs into account [23-25]. This way, study characteristics that may be deemed important can be mapped and discussed [24]. This scoping review followed the PRISMA guidelines extension for scoping reviews (PRISMA-ScR) [26].

Search strategy

To identify eligible studies, the databases of Embase, Medline, PubMed, Web of Science and PsycInfo were electronically searched for publications issued between 1 January 2000 and 7 February 2020. The search strategy was developed in collaboration with a medical research librarian and consisted of a combination of four concepts: intellectual disabilities, prevalence, chronic diseases, and comparison with the general population. Both broad (e.g. 'chronic diseases') and specific (e.g. 'diabetes mellitus') terms were used in order to ensure that all relevant studies were included in the search results. A complete overview of the search strategy is provided in Supplementary Table S1.

Study selection

Studies were included if they:

- were written in the English language;
- reported original data;
- were published in peer-reviewed journals;
- discussed the prevalence of at least IHD, CVD, DM, or COPD;
- addressed the prevalence within (a subgroup of) people with intellectual disabilities compared to people without intellectual disabilities;
- used a data collection method that was identical for people with and without intellectual disabilities.

Studies were excluded if they focused solely on conditions where intellectual disabilities cannot be assumed (i.e. cerebral palsy, autism spectrum disorder); assessed the prevalence of chronic conditions after certain interventions; focused on children only (aged 18 or below); and took place in secondary care settings only (such as hospitals or specialist care).

The initial search was conducted by the first author (MvdB), with the second author (MC) screening a random sample of 10% of all titles and abstracts. Next, the remaining articles were screened full-text by the first and the second author to assess eligibility. Disagreements were solved by discussion.

Methodological quality assessment

To better judge the results of included studies, all studies deemed eligible for inclusion were evaluated on methodological quality to assess risk of bias. The appraisal tool used – Joanna Briggs Institute Prevalence Critical Appraisal Tool – was created specifically to evaluate studies reporting prevalence data [27]. The checklist consisted of nine questions and addressed the following issues: sampling, sample size, (non)response rates, description of study participants and country, appropriate statistical analysis, and valid and reliable methods to identify the condition of intellectual disabilities. The first and the second author assessed the studies separately and later reached agreement by discussion.

The results of the quality appraisal checklist were combined into four main topics in order to provide a more structured overview. First, the findings regarding the sample were summarised; this concerned issues such as representativeness, sampling methods, and sample sizes. Second, attention was paid to the method of identification of people with intellectual disabilities. Possible influencing factors such as the use of proxy respondents, identification of intellectual disabilities based on formal diagnosis or otherwise, and method of recruiting respondents with intellectual disabilities were taken into account. Third, the manner of identification of chronic diseases was summarised, such as diagnoses in medical records or self-reported diseases. Last, the type and detail of statistical analyses performed in each study were summarised. For each topic, studies were assessed on a 5-point scale ranging from very positive (++) to very negative (--). The assessments are presented in Supplementary Table S2.

Data extraction and calculations

All data on relevant chronic diseases were extracted from the included articles. Some studies reported chronic disease prevalence for men and women separately [28] or for age groups separately [29]. In order to achieve comparability, new prevalence

rates were calculated by determining the mean of the rates for men and women (not weighted due to unavailability of population size rates) and weighted mean of the rates for the age groups. Thus, one mean prevalence rate for the total study population was computed.

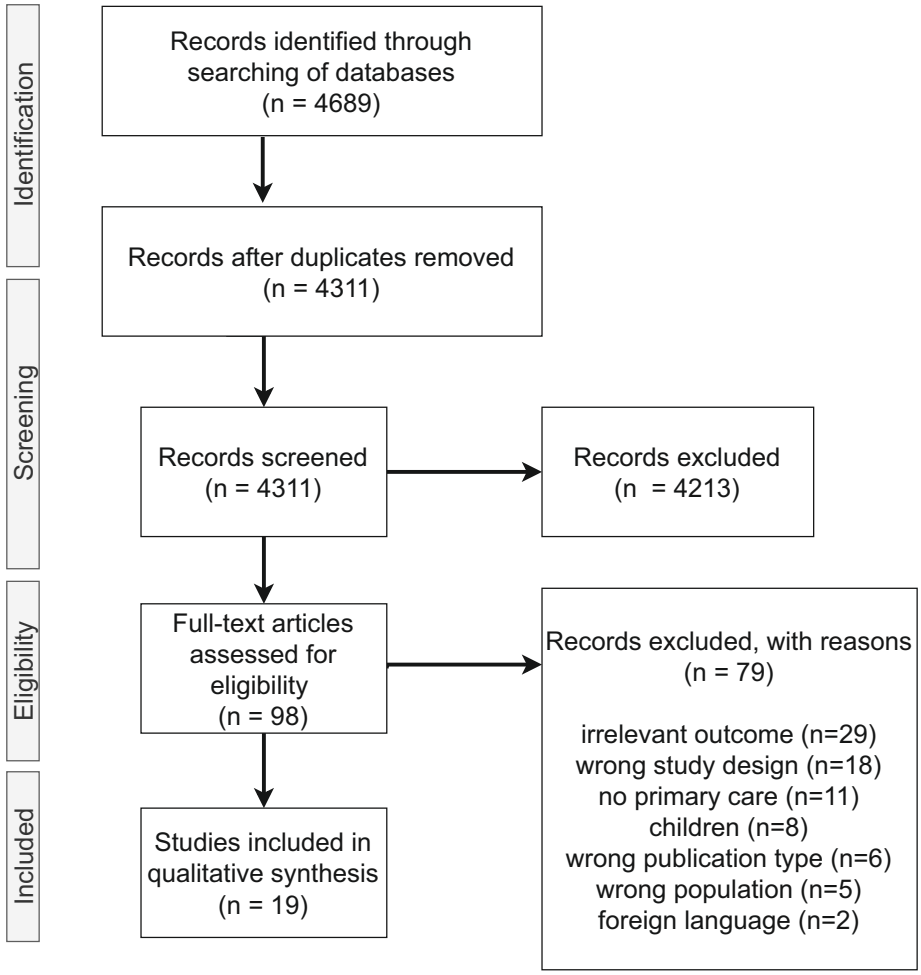

Figure 1. Search results and study selection flow chart, adapted from Moher, Liberati [30]

Characteristics of the included studies were described. First, different types of data can be used to report on chronic disease prevalence, such as register or (primary care) medical data. Next, the definition of intellectual disabilities is the way in which intellectual disabilities were operationalised in the included study. Methods for identifying someone as having intellectual disabilities consisted of a medical record of a diagnosis of intellectual disabilities,

various screening methods, or information on received services or supports specifically for people with intellectual disabilities (e.g. income support programmes, social services, residential care). Country was defined by the country in which the studies were performed, along with their dominant lifestyle and health policies, and their organisation of healthcare. Next, age groups were the ages of the included study groups that were taken into account. Lastly, sample size was the size of the group of people with intellectual disabilities and the comparison group.

RESULTS

The initial search resulted in 4,311 papers, excluding duplicates. After title and abstract screening, 98 articles were assessed full-text. There was disagreement on 14% of the articles (n=14), on which consensus was reached by discussion. This resulted in 19 studies meeting the inclusion criteria (Figure 1; [4-7, 28, 29, 31-43]). A complete overview of study characteristics and prevalence rates is shown in Table 1. Country, time period, type of data, and characteristics of the study groups are described per study. In this table, prevalence rates in percentages and the odds ratio or other reported calculations are also presented. DM was reported most often (n=18), followed by IHD (n=10), CVD (n=10), and COPD (n=8).

Characteristics of the included studies

The results of the quality appraisal are depicted in Supplementary File S2. Eight studies received a high appraisal (++ or +), eight a medium appraisal (+/-), three a low appraisal (-). The characteristics of the included studies are described in Table 1. The majority of the included studies (n=14/19) used register or (primary care) medical data to report on chronic disease prevalence, such as medical records or national patient registries. Definition of intellectual disabilities varied across studies, but most based operationalisations on ICD-9 or ICD-10 codes (n=9/19). Often, a diagnosis of intellectual disabilities was combined with diagnoses of other conditions, such as autism spectrum disorder (n=8), cerebral palsy (n=6), or foetal alcohol syndrome (n=3). The majority of the studies (n=11/19) identified people with intellectual disabilities through a diagnosis in medical records or through records of services received (n=5/19). Most studies were performed in Western-Europe (n=8/19). Three studies did not report their country, but were assumed to be performed in the USA based on earlier similar work [28, 35, 36]. In total, seven studies were performed in the USA. Most included studies took into account adults aged 18 years or older (n=11/19), others focused on adults aged 40 or 50 years and older or all ages. Lastly, the sample size across studies varied from 78 to 153,993 people with intellectual disabilities, and from 187 to 33,322,790 people without intellectual disabilities.

Table 1. Study and population characteristics of included studies on the prevalence of chronic diseases in people with and without intellectual disabilities

Source	Country (time period)	Type of data	Definition of ID
Carey, Shah [31]	England (2012)	Primary care database	QOF codes for learning disabilities, no further distinction
Cooper, Hughes-McCormack [4]	Area of Greater Glasgow, Scotland (2007–2010)	Primary healthcare register of people with intellectual disabilities	Intellectual disabilities measured by Vineland Scale in levels mild, moderate, severe, profound, and Down syndrome
Cooper, McLean [32]	Scotland (2007)	Primary care database	A set of Read Codes based on definitions used by NHS Scotland Information Services and from QOF
Dias, Ware [33]	Correctional centres in Queensland, Australia (2008–2010)	Structured questionnaire in confidential interviews	Screening with Hayes Ability Screening Index (HASI); HASI-score <85 and attended special school or received diagnosis

Characteristics of study groups		Chronic disease	Prevalence of chronic disease in n (%)		PR, RR, OR with 95% CI (ID vs. no ID)
People with ID	People without ID		People with ID	People without ID	
18+, registered in primary care; n=14,751	Matched controls; n=86,221	Ischaemic heart disease	244 (1.7)	2,316 (2.7)	PR: 0.65 (0.57-0.74)§
		Stroke and TIA	267 (1.8)	944 (1.1)	PR: 1.74 (1.52-1.98)‡
		DM	1,107 (6.9)	3,786 (4.4)	PR: 1.64 (1.53-1.75)‡
		COPD	160 (1.1)	1,184 (1.4)	PR: 0.84 (0.71-0.99)§
18+, registered in primary care; n=721	2006/7 QOF data for all adult patients within the area; n=764,672	Coronary heart disease	25 (3.5)	34,711 (4.5)	RR (rate ratio): 0.76 (0.52-1.13)†
		Stroke	13 (1.8)	15,008 (2.0)	RR: 0.92 (0.53-1.58)†
		DM	46 (6.4)	25,944 (3.4)	RR: 1.88 (1.41-2.51)***
		COPD	9 (1.2)	16,858 (2.2)	RR: 0.57 (0.29-1.09)†
People with intellectual disabilities (level not reported) aged 18+; n=8,014	People without intellectual disabilities aged 18+; n=1,416,364	Coronary heart disease	160 (2.0)	81,307 (5.7)	OR: 0.43 (0.37-0.51)***
		Stroke or TIA	171 (2.1)	36,374 (2.6)	OR: 1.19 (1.02-1.37)†
		DM	531 (6.6)	74,300 (5.3)	OR: 1.63 (1.49-1.79)***
		COPD	209 (2.6)	52,898 (3.7)	OR: 0.84 (0.73-0.97)***
Prisoners; n=115	Prisoners, screened with HASI, score >85; n=1164	DM	6 (5.4)	63 (5.1)	aOR: 1.3 (0.5-3.3)†

Table 1. Continued

Source	Country (time period)	Type of data	Definition of ID
Durbin, Jung [34]	Ontario, Canada (2010)	Health administrative databases	Disabilities income support programmes and algorithm that uses information from diagnostic codes (intellectual disabilities, foetal alcohol syndrome, autism spectrum disorder, other pervasive developmental disorders, chromosomal and autosomal anomalies)
Erickson and Kornexl [35]	Mid-western academic medical centre (2011)	Registration data	ICD-9 diagnosis of mental retardation or having diagnosis of one of the more common conditions associated with developmental disabilities (autism spectrum disorder, Down syndrome, Williams syndrome, fragile X syndrome, cerebral palsy, foetal alcohol syndrome)
Erickson, Spoutz [36]	Mid-western academic medical centre (2012)	Medical records	ICD-9 diagnosis of condition related to intellectual disabilities: Down syndrome, foetal alcohol syndrome, cerebral palsy, autism spectrum disorder, mental retardation, developmental disabilities, not specified/ other

Characteristics of study groups		Chronic disease	Prevalence of chronic disease in n (%)		PR, RR, OR with 95% CI (ID vs. no ID)
People with ID	People without ID		People with ID	People without ID	
Newcomers aged 19-65; n=2,830	Newcomers aged 19-65; n=1,646,803	DM	280 (9.9)	119,768 (7.3)	aPR/RR: 1.97 (1.77-2.20)§
		COPD	67 (2.4)	28,343 (1.7)	aPR/RR: 2.11 (1.68-2.66)§
18+, patient in general internal medicine practice; n=183	18+, patient in general internal medicine practice; n=497	Myocardial infarction	3 (1.6)	2 (0.4)†	
		Stroke	10 (5.5)	7 (1.4)**	
		DM	19 (10.4)	74 (14.9)†	
People with intellectual and developmental disabilities aged 40-79 years without a history of cardiovascular disease; n=78	People without intellectual and developmental disabilities aged 40-79 years without a history of cardiovascular disease; n=187	Myocardial infarction (40-years)	1 (2.3)	0 (0)†	
		Stroke (40-years)	2 (4.7)	0 (0)†	
		DM (40- years)	4 (9.3)	4 (3.1)†	
		DM (40-79 years)	9 (11.5)	36 (19.3)†	

Table 1. Continued

Source	Country (time period)	Type of data	Definition of ID
Flygare Wallen, Ljunggren [29]	Stockholm, Sweden (2010)	Administrative data on healthcare	ICD-10 diagnosis of moderate, severe, profound, other or unspecified intellectual disabilities, unspecified disorder of psychological development, Down syndrome, trisomy 18, trisomy 13, fragile X syndrome, congenital malformation syndromes, Rett's syndrome, autism spectrum disorder, other childhood disintegrative disorder, Asperger's syndrome, other pervasive developmental disorders
Haider, Ansari [37]	Victoria, Australia	Survey (general population), administrative database (ID)	N.R.
Havercamp, Scandlin [38]	North Carolina, USA (2001)	Survey (general population), registration data and interviews (intellectual disabilities)	Random sample of adults with developmental disabilities receiving special services; self-reported developmental disabilities
Hedgeman, Ulrichsen [39]	Denmark (1995–2012)	Danish National Patient Registry	Prader-Willi syndrome, diagnosis made in study period by ICD-code of DQ871E

Characteristics of study groups		Chronic disease	Prevalence of chronic disease in n (%)		PR, RR, OR with 95% CI (ID vs. no ID)
People with ID	People without ID		People with ID	People without ID	
Persons with intellectual disabilities excluding Down syndrome; n=11,785; Persons with Down syndrome; n=1,282	Persons without any diagnosis of intellectual disabilities, Down syndrome, or autism spectrum disorder; n=1,996,140	DM (18+, women, intellectual disability vs no intellectual disability)	251 (8.2)	50,171 (6.3)	OR: 2.40 (2.11-2.73)‡
		DM (18+, men, intellectual disability vs no intellectual disability)	342 (9.0)	64,621 (8.5)	OR: 2.01 (1.80-2.24)‡
		DM (18+, women, Down syndrome vs no intellectual disability)	19 (5.5)	50,171 (6.3)	OR: 1.78 (1.17-2.73)‡
		DM (18+, men, Down syndrome vs no intellectual disability)	15 (3.9)	64,621 (8.5)	OR: 0.70 (0.42-1.17)†
Proxy respondents on behalf of people with intellectual disabilities; n=897	n=34,168	Stroke	N.R. (2.0)	N.R. (2.5)†	
		DM	N.R. (8.9)	N.R. (5.8)*	
Information obtained via registration/ medical data, interviews with person or proxy respondent; n=946	Two groups: No disabilities (n=4,358), Disabilities (n=1,598)	DM (intellectual disabilities vs no disabilities)	N.R. (3.9)	N.R. (7.9)	RR: 2.0 (1.4-2.9)*
All persons with Prader-Willi syndrome n=155	General population; n=15,500	Myocardial infarction	x	31 (0.2)§	
		DM	14 (9.0)	162 (1.0)§	

Table 1. Continued

Source	Country (time period)	Type of data	Definition of ID
Jansen, Rozeboom [40]	Two Dutch care providers for people with ID aged 50+ (2007)	Medical records of general practice patients in two Dutch care providers and primary healthcare in same region	Indication for residential care and specialist primary healthcare, based on mild, moderate, severe, profound intellectual disabilities, Down syndrome
McCarron, Cleary [5]	Ireland (2010)	Cohort study (incl. in-person interviews, questionnaire, and physical health assessment)	Receiving services
McDermott, Moran [28]	Country not reported (1990–2003)	Medical records	ICD-9 diagnosis of autism spectrum disorder, cerebral palsy with and without intellectual disabilities, psychiatric disabilities with intellectual disabilities, other intellectual disabilities
McDermott, Moran [41]	South Carolina, USA (1990–2003)	Medical records	ICD-9 diagnosis of autism spectrum disorder, cerebral palsy with and without mental retardation, psychiatric disabilities with mental retardation, other mental retardation
McDermott, Moran [42]	South Carolina, USA (1990–2003)	Medical records	ICD-9 diagnosis of autism spectrum disorder, cerebral palsy with and without mental retardation, psychiatric disabilities with mental retardation, other mental retardation

Characteristics of study groups		Chronic disease	Prevalence of chronic disease in n (%)		PR, RR, OR with 95% CI (ID vs. no ID)
People with ID	People without ID		People with ID	People without ID	
Individuals aged 50 and over; n=510	All patients aged 50 years and over registered in a large general practice in the same area; n=823	Lifetime prevalence of CVD and/ or myocardial infarction	25 (5.7)	36 (4.4)†	
Adults aged 50 years or older in intellectual disabilities database; n=478	Adults aged 50 years or older; n=478	Heart attack	7 (1.5)	15 (3.1)†	
		DM	52 (11.1)	31 (6.5)*	
Adults with intellectual disabilities in primary care medical practices; n=652	Matched patients based on age at entry into general practice; n=1,828	DM (total intellectual disabilities group)	61 (9.8)	265 (14.5)	OR: 1.1 (0.8-2.2)†
		DM (only intellectual disabilities)	82 (12.7)	265 (14.5)	OR: 1.4 (0.9-2.1)†
Adults with developmental disabilities condition registered in primary care; n=692	Matched patients based on age at entry into general practice; n=2,084	TIA	N.R. (0.3)	N.R. (1.7)§	
		DM	N.R. (10.4)	N.R. (15.8)§	
		COPD	N.R. (6.4)	N.R. (9.5)§	
Adults with developmental disabilities condition registered in primary care; n=692	Matched patients based on age at entry into general practice; n=2,084	TIA (women)	N.R. (0.0)	N.R. (1.7)	
		TIA (men)	N.R. (0.5)	N.R. (1.4)	HR: 0.96 (0.64-1.45)†
		DM (women)	N.R. (12.3)	N.R. (16.2)	
		DM (men)	N.R. (8.7)	N.R. (13.8)	HR: 1.04 (0.57-1.89)†
		COPD (women)	N.R. (4.6)	N.R. (8.6)	
		COPD (men)	N.R. (7.9)	N.R. (10.2)	

Table 1. Continued

Source	Country (time period)	Type of data	Definition of ID
Morin, Mérineau-Côte [7]	Quebec, Canada (2010)	Province-wide mail survey	Receiving services from an agency for intellectual disabilities and autism spectrum disorder or from social services (eligibility based on AAIIDD definition)

Characteristics of study groups		Chronic disease	Prevalence of chronic disease in n (%)		PR, RR, OR with 95% CI (ID vs. no ID)
People with ID	People without ID		People with ID	People without ID	
Individuals aged 15 years and older receiving services; n=789	People aged 15 years and older; n not reported	DM (intellectual disabilities vs no intellectual disabilities)	N.R. (8.3)	N.R. (5.1)†	
		DM (mild/ moderate intellectual disabilities vs no intellectual disabilities)	N.R. (8.6)	N.R. (6.4)†	
		DM (severe/ profound intellectual disabilities vs no intellectual disabilities)	N.R. (4.8)	N.R. (6.4)†	
		DM (Down syndrome vs no intellectual disabilities)	N.R. (4.2)	N.R. (5.1)†	

Table 1. Continued

Source	Country (time period)	Type of data	Definition of ID
Perera, Audi [43]	Haringey and London, England (2016–2017)	Health and social care register	Diagnosis of learning disabilities in medical record (from learning disabilities register)
Tyler, Schramm [6]	Cleveland, USA (2005–2008)	Electronic health records	ICD-9 diagnosis of one of the following: intellectual disabilities, cerebral palsy, chromosomal abnormalities (incl. Down syndrome), pervasive developmental disorders (incl. autism spectrum disorder), unspecified delay in development, anomalies of the brain

† = no significant difference; ‡ = significant difference, p-level not reported; * = $p<0.05$; ** = $p<0.01$; *** = $p<0.001$; § = significance not reported.

ID = intellectual disabilities; IHD = ischemic heart disease; CVD = cerebrovascular disease;

Characteristics of study groups		Chronic disease	Prevalence of chronic disease in n (%)		PR, RR, OR with 95% CI (ID vs. no ID)
People with ID	People without ID		People with ID	People without ID	
All persons aged 0+ registered in general practice in England; n=1,078 (Haringey), n=28,078 (London), n=153,993 (total England)	All persons registered in general practice in England; n=282,478 (Haringey), n=7,559,949 (London), n=33,322,790 (total England)	Coronary heart disease (Haringey (H))	N.R. (0.7)	N.R. (1.6)§	
		Coronary heart disease (London (L))	N.R. (0.9)	N.R. (0.0)§	
		Coronary heart disease (total England (E))	N.R. (1.1)	N.R. (3.1)§	
		Stroke or TIA (H)	N.R. (1.1)	N.R. (0.9)§	
		Stroke or TIA (L)	N.R. (1.5)	N.R. (1.1)§	
		Stroke or TIA (E)	N.R. (1.7)	N.R. (1.7)§	
		DM type 1 (H)	N.R. (0.5)	N.R. (0.2)§	
		DM type 1 (L)	N.R. (0.6)	N.R. (0.3)§	
		DM type 1 (E)	N.R. (0.7)	N.R. (0.4)§	
		COPD (H)	N.R. (1.1)	N.R. (0.9)§	
		COPD (L)	N.R. (1.0)	N.R. (1.2)§	
		COPD (E)	N.R. (1.1)	N.R. (1.9)§	
Persons of 18 years or older receiving ongoing healthcare at the Cleveland Clinic; n=1,267	One-to-one match by age, sex, race, and health insurance status with two other patients who received similar care during the same study period; n=2,534	Coronary heart disease	33 (3.5)	196 (7.7)	0.43 (0.31-0.60)***
		DM	131 (10.3)	384 (15.2)	0.65 (0.52-0.80)***
		COPD	41 (3.2)	145 (5.7)	0.55 (0.39-0.78)***

DM = diabetes mellitus; COPD = chronic obstructive pulmonary disease; TIA = transient ischemic attack; (a)PR = (adjusted) prevalence risk; (a)OR = (adjusted) odds ratio; RR = relative risk (unless stated otherwise); CI = confidence interval; x = size too low to report (1–5 observations); N.R. = not reported.

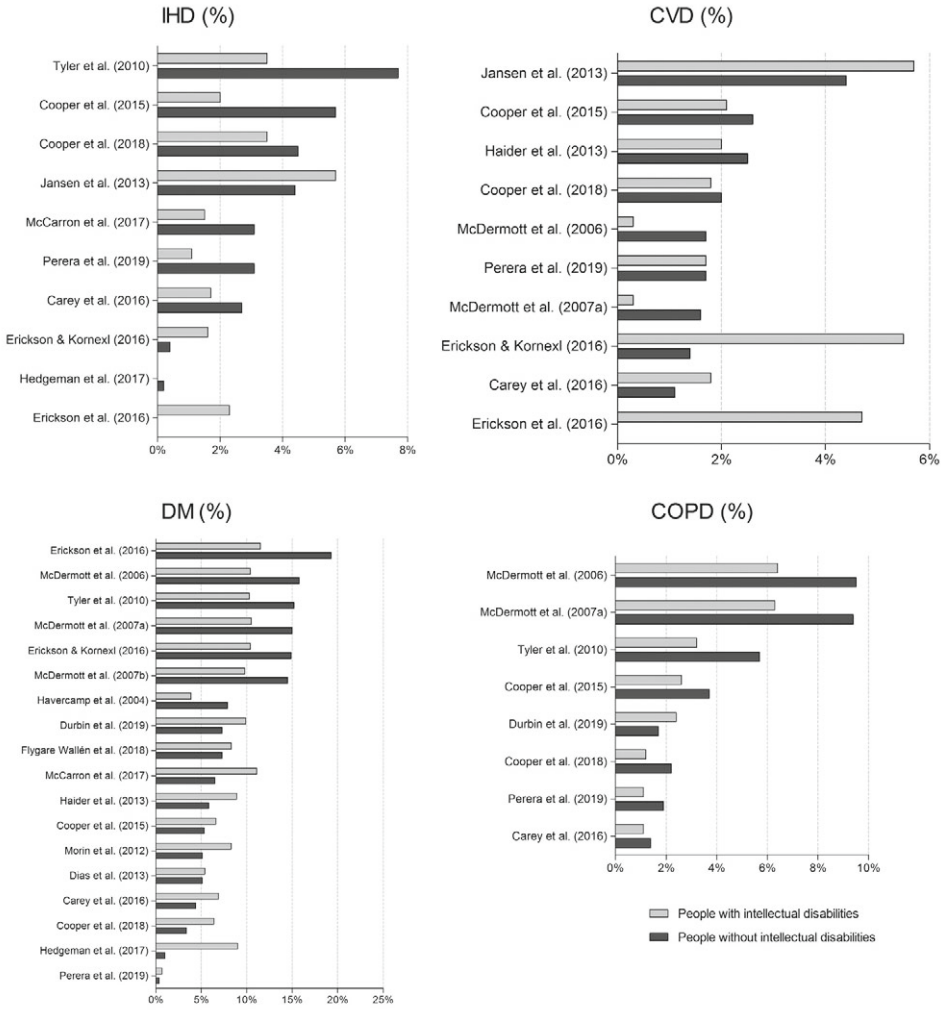

Figure 2. Prevalence of ischaemic heart disease (IHD), cerebrovascular disease (CVD), diabetes mellitus (DM), and COPD

IHD prevalence

Studies (n=10/19) reported IHD prevalence rates between 0.0% and 5.7% for people with intellectual disabilities, and 0.0% to 7.7% for people without intellectual disabilities (Figure 2). In most studies, IHD prevalence was lower for people with intellectual disabilities compared to people without intellectual disabilities. One study that stratified by severity levels of intellectual disabilities reported higher IHD prevalence in more severe levels [40]. The highest IHD prevalence rates among people with and without intellectual disabilities were found among the studies with a high-quality appraisal [6, 40] (Table 2). The range in IHD prevalence was higher in

studies where the population of people with intellectual disabilities was identified through relevant diagnoses in medical records rather than through other methods (Figure 3). The two studies identifying intellectual disabilities through support or services both focused on adults aged 50 years or older [5, 40], of which one shows highest IHD prevalence among people with intellectual disabilities [40]. In studies performed in the USA, IHD prevalence had the highest range for people without intellectual disabilities compared to other countries (Figure 4). Studies performed in Great Britain utilised larger samples, which likely contributed to lower IHD prevalence compared to other countries.

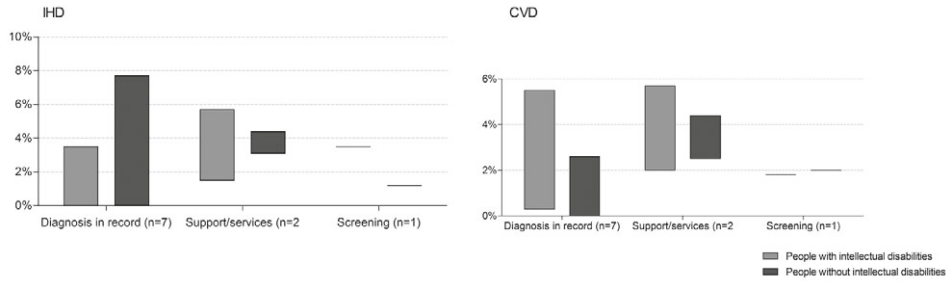

Figure 3. Range in ischemic heart disease (IHD) and cerebrovascular disease (CVD) prevalence (%) in the literature, split by type of identification of intellectual disabilities in data

CVD prevalence

CVD prevalence in the included studies (n=10/19) varied from 0.3% to 5.7% among people with intellectual disabilities, and from 0.0% to 4.4% among people without intellectual disabilities (Figure 2). One study reported prevalence by severity levels: the higher the severity level of intellectual disabilities, the higher the CVD prevalence [40]. The range in prevalence among people with intellectual disabilities was higher when diagnoses of intellectual disabilities in medical records were used as the indicator (Figure 3). The USA had the highest range in CVD prevalence among people with intellectual disabilities. In the UK, the range in CVD prevalence was higher among people without intellectual disabilities (Figure 4). The highest CVD prevalence among people both with and without intellectual disabilities was reported by a study including adults aged 50 years and older [40]; the lowest prevalence rates were reported by Erickson, Spoutz [36] who included ages 40 years or less (Table 2). The highest difference in prevalence rates between people with and without intellectual disabilities could be found among the study using the smallest samples [36]. Studies performed in Great Britain in general utilised larger samples compared to other countries.

Table 2. Summary of patterns in study and population characteristics across prevalence studies

	Ischaemic heart disease	Cerebrovascular disease
Quality appraisal	Highest prevalence rates in studies with highest appraisal	No pattern
Type of data	No pattern	No pattern
Definition of intellectual disabilities	No pattern	No pattern
Method of identification of intellectual disabilities	Higher prevalence in studies using received support/services compared to diagnoses in medical records	Highest prevalence among studies using received support/services compared to other measurements
Country	In UK and Ireland, higher prevalence in general population compared to population with intellectual disabilities, in USA other way around	Highest range of prevalence among population with intellectual disabilities in USA, in UK the smallest
Age groups	No pattern	Highest prevalence rates in study focusing on elderly (50+ years), lowest among study focusing on younger persons (40- years)
Sample size	No pattern	Most difference in prevalence rates among study using smallest samples

	Diabetes mellitus	Chronic obstructive pulmonary disease
	Highest prevalence rates in general population in studies with negative appraisal	Highest prevalence rates in general population and population with intellectual disabilities in medium appraisal studies
	No pattern	No pattern
	No pattern	No pattern
	Highest and lowest prevalence in general population and population with intellectual disabilities among studies using intellectual disabilities-related diagnoses in medical records	No pattern
	In USA, population with intellectual disabilities has higher prevalence rates compared to general population, in other countries vice versa	Highest prevalence among general population and population with intellectual disabilities in USA, relatively low prevalence in UK and Canada
	Studies focusing on all ages present lowest prevalence rates in general population and highest prevalence in population with intellectual disabilities	Lowest prevalence in study focusing on all ages
	Highest prevalence rates in general population and population with intellectual disabilities in smaller samples, lowest prevalence rates in larger samples	Lower prevalence rates in studies with larger sample sizes, highest prevalence in smallest samples

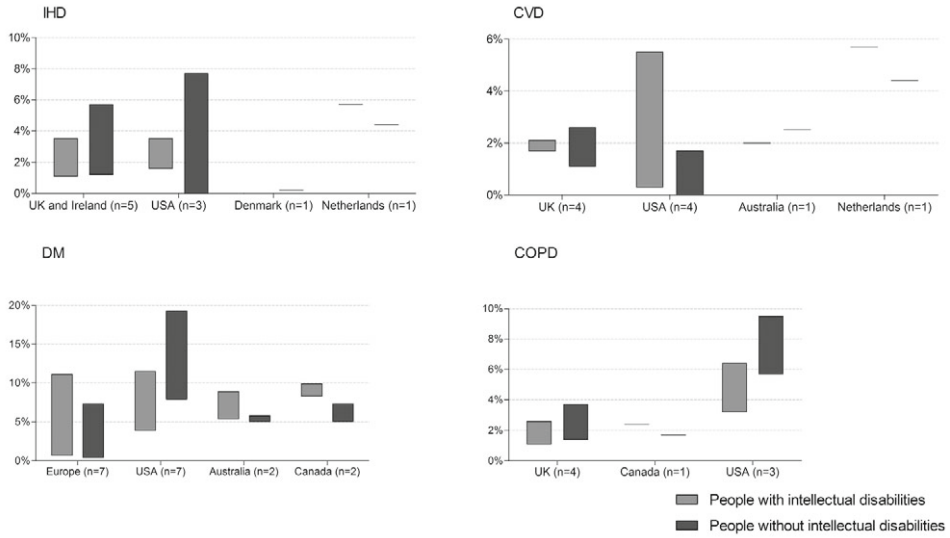

Figure 4. Range in chronic disease prevalence (%) in the literature by country

DM prevalence

The prevalence of DM varied in studies (n=18/19) from 0.7% to 11.5% among people with intellectual disabilities, and from 0.4% to 19.3% among people without intellectual disabilities (Figure 2). DM prevalence was mostly higher for people with intellectual disabilities than for people without intellectual disabilities, except in studies that found high prevalence rates among people without intellectual disabilities (>10%). Only two studies distinguished between type 1 and 2 diabetes [41, 42]. Both the highest and the lowest DM prevalence for people with and without intellectual disabilities were found in studies using diagnoses related to intellectual disabilities in medical records (Table 2). DM prevalence among people with intellectual disabilities was generally lower in the USA compared to those without intellectual disabilities, whereas the opposite was true for Western-Europe (Figure 4). The studies with highest appraisal were performed in Western-Europe [32, 39]. The two studies focusing on all ages reported the highest prevalence among people with intellectual disabilities and the lowest DM prevalence among people without intellectual disabilities [39, 43]. Lastly, the smallest sample size corresponds with the highest DM prevalence in people both with and without intellectual disabilities [36], whereas the lowest prevalence rates can be found in the largest sample size [43]. The highest DM prevalence among people with and without intellectual disabilities was reported in a study from the USA with smallest sample, which focused on the oldest age groups (40-79 years) compared to the other studies [36].

COPD prevalence

Studies on COPD (n=8/19) reported prevalence rates from 1.1% to 6.4% among people with intellectual disabilities, and from 1.4% to 9.5% among people without intellectual disabilities (Figure 2). In all but one study (Durbin, Jung [34], the prevalence of COPD was lower in people with intellectual disabilities compared to people without intellectual disabilities. The highest COPD prevalence was reported by two studies with a medium appraisal [41, 42]. COPD prevalence was highest in the USA compared to studies performed in other countries, and showed the largest differences between people with and without intellectual disabilities (Figure 4). Prevalence rates in the UK were more comparable between people with and without intellectual disabilities, and overall lowest across the included studies. The only study considering all ages reported the lowest COPD prevalence [43] (Table 2). A larger sample size was accompanied by a lower COPD prevalence [43], a smaller sample size by a higher prevalence [41, 42].

DISCUSSION

This scoping review is the first to map the broadness of published literature on chronic disease prevalence in people with intellectual disabilities compared to people without intellectual disabilities. Chronic disease prevalence varied considerably between studies and differed when study characteristics were taken into account. This study builds upon existing chronic disease prevalence reviews by exploring their observations that methodological differences in the included studies could possibly be important in explaining variances in prevalence rates. The reviews mention methodological differences such as operational definition and method of identification of intellectual disabilities, differences in study groups in terms of sex and aetiology of intellectual disabilities, method of data collection, sample size, and method of diagnosis of chronic diseases [1, 2, 44, 45]. Other similar reviews either did not take the role of methodological choices into account or focused on different health problems [44, 46]. This study is therefore the first to offer guidance to primary care providers and researchers in interpreting chronic disease prevalence in people with intellectual disabilities.

This review described characteristics of included studies and identified five valuable aspects that are important when interpreting chronic disease prevalence in people with intellectual disabilities; being type of data, identifying of intellectual disabilities, country, age of the study groups, and sample size. These aspects are discussed one by one: First, when interpreting results, one should always be aware of the consequences of different types of data. Studies relying on self-reported values are at risk of potential bias, which may result in an over- or underestimation of a person's ill-health. In people

with intellectual disabilities, self-reporting can be accompanied by extra challenges [47], and therefore studies often resort to using proxy respondents. However, proxy reporting decreases the validity of the results [48, 49] and complicates comparison between people with and without intellectual disabilities.

Second, this study emphasises the value of recognising the way in which intellectual disabilities are identified and defined across studies. Although most included studies used similar methods for identifying intellectual disabilities (via medical records or records of specific services), chronic disease prevalence was still diverse in these studies. This finding suggests that studies using the same methods for identifying people with intellectual disabilities do not necessarily include the same populations, as people with intellectual disabilities are identifiable via multiple sources. Earlier research supports the finding that using different identification methods as well as different definitions of intellectual disabilities may complicate estimating prevalence rates [50].

Only a few countries have national registers from which intellectual disabilities can be identified in a relatively reliable manner; other methods are often less conclusive [51]. Frequently, many different conditions related to intellectual disabilities were examined simultaneously, but in conditions such as autism or cerebral palsy intellectual disabilities cannot always be assumed [52, 53].

Third, the country in which studies were performed was relevant for interpreting chronic disease prevalence. Interestingly, in the USA, the prevalence of cardiovascular diseases (IHD and CVD) was consistently higher among people with intellectual disabilities compared to people without intellectual disabilities, whereas COPD and DM in the USA were more prevalent among people without intellectual disabilities. Prevalence of IHD, CVD, DM, and COPD was high in the USA among people both with and without intellectual disabilities compared to other countries. A possible explanation is the higher prevalence of unhealthy lifestyles, and consequently obesity levels, in the USA [54], given that these diseases are all related to unhealthy lifestyles [55, 56]. In addition, some argue that American health promotion policies can be prone to reinforce health inequalities [57], whereas European policies seem more inclusive [58]. Furthermore, the differences in primary care systems in the USA and European countries can result in different timings in diagnosis and management of chronic diseases [59, 60]. When interpreting and comparing health statuses of people with intellectual disabilities residing in the USA and Western-Europe these differences should therefore always be kept in mind.

Fourth, the role of age should always be noted in studies on chronic disease prevalence. Although the life expectancy of people with intellectual disabilities has increased, they often show earlier signs of aging compared with people without intellectual disabilities [61], resulting in higher mortality rates [62]. Results and comparability between people with and without intellectual disabilities can be affected by this earlier aging effect, as the occurrence of chronic diseases is generally higher with increasing age [63, 64], and as several chronic diseases are more common among aging people with intellectual disabilities than among aging people without intellectual disabilities [65]. In line with these previous findings, this review found that studies only taking older age groups into account were more likely to report higher prevalence of chronic diseases in people with intellectual disabilities.

Fifth, sample sizes should be critically evaluated when one is interpreting differences in prevalence rates of chronic diseases. In the case of COPD and DM, it could be seen that a higher sample size was accompanied by a lower prevalence, and vice versa. This can be explained by the fact that larger sample sizes are generally better suited to make more precise claims and are more likely to have included a representative sample [66].

Strengths and limitations

This review has some limitations. First, we restricted our scope of chronic disease to IHD, CVD, DM, and COPD. Diseases that are more prevalent among people with intellectual disabilities, for instance epilepsy [67] or chronic skin disease [68], were not taken into account. We chose to focus on the four most prevalent types of chronic conditions that have a large global impact as well as a high impact on the everyday lives of people with intellectual disabilities. Second, few studies included in this review make necessary distinctions, such as between diabetes type 1 and type 2, ischaemic or haemorrhagic stroke, or severity levels of intellectual disabilities. However, diabetes type 1 and type 2 have different manifestations and aetiology [69]. Not being able to make these distinctions complicates the formulation of adequate disease management methods for specific diseases.

Notwithstanding these limitations, this review provides the first exploration of literature on chronic disease prevalence rates in people with intellectual disabilities compared to people without intellectual disabilities. Although Jansen et al. conducted a similar review in 2004 [44], they focused solely on the prevalence of several health problems that were not included in this review, such as epilepsy and sensory loss. The current review is in line with another review that explored how methodological choices may influence multimorbidity prevalence rates [46]. Comparable to the current review, the authors concluded that type of data, country, and age groups are important in assessing multimorbidity in the general population. However, intellectual disabilities

were not taken into account [46]. This review therefore offers direction in interpreting studies on chronic disease prevalence in people with intellectual disabilities. Second, it offers a first insight into the comparative health regarding chronic diseases of people with intellectual disabilities compared to the general population. Third, a large variety of studies have been taken into account. Although study characteristics such as age or sex are better known influences on prevalence rates [29, 43], this review highlights the significance of other, less often examined characteristics, such as type of data. In traditional reviews, the great heterogeneity in study designs, populations, and countries is associated with challenges in summarising evidence, but by performing a scoping review it was possible to explore such characteristics in greater depth. Fourth, the fact that we were able to perform a quality assessment increases the legitimacy of the claims made.

Recommendations for future research

This review provides a fruitful basis upon which to build future research on chronic diseases in people with intellectual disabilities. First, as the current review is the first to explore the role of study designs, populations, and countries in chronic disease prevalence, this study can be used as a valuable basis for conducting further research, such as a meta-analysis. In addition, no studies conducted in non-Western countries were identified. Research demonstrates that chronic diseases represent a high burden in non-Western, low- or middle-income, or less developed countries [70, 71]. The situation of people with intellectual disabilities is also very different in such countries, but this global difference is not often studied [72]. The prevalence rates of IHD, CVD, DM, and COPD as presented in this review are therefore a representation of Western countries. Next to the use of different methods or countries, this review has also identified several important aspects that future research should take into account when both studying and interpreting chronic disease prevalence in people with intellectual disabilities. First, future research should disclose as much as possible the study and population characteristics. Existing guidelines for prevalence studies, such as STROBE or RECORD [73, 74], are useful tools and should be utilised widely. This way, the need for valid and reliable information on the health of people with intellectual disabilities [75] can be better met. Second, in order to make useful claims future studies on chronic disease prevalence should take into account multiple interacting factors, such as age [36, 40, 43] or sex [29, 42], but also factors such as type of data or identification of intellectual disabilities. Additionally, future research should report chronic disease prevalence by severity levels of intellectual disabilities if possible. The few studies that do so report possibly important patterns in chronic diseases [4, 40, 76]. Third, large population studies should be conducted in order to obtain reliable and valid prevalence estimates. In this type of study, entire populations can be taken into account, resulting

in thoroughly defined and representative study populations [77]. Because it currently still is difficult to identify people with intellectual disabilities in population datasets [49], future studies should be transparent in the methods used to identify people with intellectual disabilities.

Lastly, comparisons between incidence and prevalence rates can prove interesting research subjects. While prevalence rates are useful for indicating disease burden, incidence rates give insight in the occurrence rate of chronic diseases in populations [78].

Conclusion

This review adds to the literature by providing a first exploration of the broadness of published literature on chronic disease prevalence in people with intellectual disabilities and by describing main characteristics of these studies. Chronic disease prevalence varies greatly between people with and without intellectual disabilities across studies. Although study characteristics such as country and age group are more apparent influencers in chronic disease prevalence, this review also highlights the importance of other factors that are less often examined, such as type of data and definition of intellectual disabilities. Researchers should therefore acknowledge the influence of study characteristics and methodologies when studying chronic disease prevalence in people with intellectual disabilities. This review underscores the need for transparent and comparable prevalence studies. The great heterogeneity in study characteristics and methodologies complicate generalisation of study results. Rather, this review argues that prevalence rates should always be interpreted in the context of methodology. Only then, primary care providers and public health planners are able to utilise prevalence rates of chronic diseases and apply them into practice.

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APPENDIX

Supplementary Table S1. Search strategy for PubMed[a]

Concept	Search terms
Intellectual disabilities	"Intellectual Disability"[Mesh] OR "Mentally Disabled Persons"[Mesh] OR "Developmental Disabilities/complications"[Mesh] OR "Developmental Disabilities"[Mesh] OR "Specific Learning Disorder"[Mesh] OR intellectual development disorder*[tiab] OR mentally challenged[tiab] OR learning disabilit*[tiab] OR special needs[tiab] OR low IQ[tiab] OR developmental disab*[tiab] OR down syndrome*[tiab] OR downs syndrome*[tiab] OR prader-willi[tiab] OR fragile X[tiab] OR Trisomy 21[tiab] OR Trisomy21[tiab] OR trisomies[tiab] OR mongolism*[tiab]
Prevalence	"Prevalence"[Mesh] OR "Epidemiology"[Mesh] OR epidemiology[MeSH Subheading] OR "Cross-sectional studies"[Mesh] OR "Incidence"[Mesh] OR prevalence[tiab] OR epidemiology[tiab] OR incidence[tiab]
Chronic diseases	"Chronic Disease"[Mesh] OR "Healthcare Disparities"[Mesh] OR "Health Status"[Mesh] OR "Comorbidity"[Mesh] OR "Morbidity"[Mesh] OR "Multiple Chronic Conditions"[Mesh] OR chronic disease*[tiab] OR chronic illness*[tiab] OR health disparit*[tiab] OR health status*[tiab] OR comorbid*[tiab] OR morbidity[tiab] OR multimorbidity[tiab] OR chronically ill[tiab] OR "Cardiovascular Diseases" [Mesh] OR heart attack*[tiab] OR myocardial infarct*[tiab] OR heart infarct*[tiab] OR Stroke*[tiab] OR cerebrovascular[tiab] OR cva[tiab] OR Coronary artery disease*[tiab] OR coronary arteriosclerosis[tiab] OR atherosclerosis[tiab] OR Peripheral Arterial Disease*[tiab] OR Congenital heart*[tiab] OR heart abnormalit*[tiab] OR heart defect*[tiab] OR Venous thromboembolism[tiab] OR deep vein thrombosis[tiab] OR dvt[tiab] OR pulmonary embolism[tiab] OR lung embolism[tiab] OR pulmonary thromboembolism[tiab] OR lung thromboembolism[tiab] OR "Respiratory Tract Diseases"[Mesh] OR respiratory tract disease*[tiab] OR respiratory hypersensit*[tiab] OR bronchial disease*[tiab] OR Asthma[tiab] OR Asthmas[tiab] OR COPD[tiab] OR Chronic Obstructive Pulmonary Disease[tiab] OR COAD[tiab] OR Chronic Obstructive Airway Disease[tiab] OR Respiratory disease*[tiab] OR "Diabetes mellitus"[Mesh] OR diabetes[tiab]
Comparison with general population	"Matched-Pair Analysis"[Mesh] OR "Case-Control Studies"[Mesh] OR "Control Groups"[Mesh] OR "Probability"[Mesh] OR matched pair analys*[tiab] OR case-control stud*[tiab] OR control group*[tiab] OR odds ratio[tiab] OR comparison[tiab] OR compared[tiab]

[a] The search terms for Web of Science are similar, but adapted to its specific format.

Supplementary Table S2. Result of appraisal checklist

	Overall rate	Sample	Identifying intellectual disabilities
Cooper, McLean [32]	++	+ Sufficient Use of register data	+ Sufficient Use of QOF codes for learning disabilities
Hedgeman, Ulrichsen [39]	++	+ Sufficient All people with Prader-Willi syndrome in Denmark are included and matched with general population on 1:100	+ Sufficient Identification via diagnosis of intellectual disabilities in medical record
Carey, Shah [31]	+	+ Sufficient Use of register data (primary care database)	+ Sufficient Use of QOF codes for learning disabilities
Durbin, Jung [34]	+	+ Sufficient Use of register data, namely health administrative databases	+/- Debatable Identification is algorithm based, but not a validated one
Erickson, Spoutz [36]	+	+ Sufficient All available cases are taken into account, but low N in both group with and without intellectual disabilities	+/- Debatable Identification via diagnosis of intellectual disabilities in medical record, but only 1/3 of this population had an official diagnosis
Flygare Wallen, Ljunggren [29]	+	+ Sufficient Administrative data on healthcare, thus use of register data	+ Sufficient Identification via diagnosis of intellectual disabilities in medical record
Jansen, Rozeboom [40]	+	+/- Debatable Study population consists of residents from two care providers; this is not representative of the larger population	+ Sufficient Institutionalised people with intellectual disabilities are taken into account, thus use of medical records
Tyler, Schramm [6]	+	+ Sufficient Electronic health records and use of register data	+ Sufficient Identification of intellectual disabilities via diagnosis in medical record

	Identifying chronic disease	Statistical analysis
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+/- Debatable Methods were described in detail to properly identify the analytical method. Did not report significance, but did report n, %, and PR
	+ Sufficient Chronic disease(s) identified by validated algorithms	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. N and % were reported, but confidence intervals and significance levels were not
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+/- Debatable Methods were too poorly described to properly identify the analytical method. The statistical analysis was sufficient

Supplementary Table S2. Continued

	Overall rate	Sample	Identifying intellectual disabilities
Cooper, Hughes-McCormack [4]	**+/-**	- Insufficient Use of register data, but underrepresentation of intellectual disabilities (1:1000). Unclear how sampling took place for both groups. Data on age and gender were unknown for general population	+/- Debatable Identification based on screening
Erickson and Kornexl [35]	**+/-**	+/- Debatable All available cases are taken into account. A high percentage of people with intellectual disabilities is 'lost', unclear whether they do not have CVD risk factors or whether there are no data available	+/- Debatable Identification via diagnosis of intellectual disabilities in medical record, but only 1/3 of ID population had an official diagnosis
Haider, Ansari [37]	**+/-**	+ Sufficient Sample is randomly selected. There are no differences in non-responders and responders	- Insufficient No definition of intellectual disabilities is given. Only those are included who sought assistance. Proxy respondents were used. People with intellectual disabilities were invited to participate via CATI, but this is not a suitable method for this group
McCarron, Cleary [5]	**+/-**	+ Sufficient Random representative sample, no significant differences in non-responders and responders	+/- Debatable Identification of intellectual disabilities via service or support, 1/3 of these respondents are proxy respondents
McDermott, Moran [41]	**+/-**	+/- Debatable Use of medical records, unknown whether it is a random sample	+ Sufficient Identification of intellectual disabilities via diagnosis in medical record
McDermott, Moran [28]	**+/-**	+/- Debatable Use of medical records, unknown whether it is a random sample	+/- Debatable Identification of intellectual disabilities via diagnosis in medical record

	Identifying chronic disease	Statistical analysis
	- Insufficient Measurement of chronic disease is unclear for the general population	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	- Insufficient Chronic disease(s) identified by self-reported values	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. The %, confidence intervals, and significance are reported, but no N. There were few dropouts
	- Insufficient Chronic disease(s) identified by self-reported values	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. Only % was reported, no confidence intervals or significance levels
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+ Sufficient Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was sufficient

Supplementary Table S2. Continued

	Overall rate	Sample	Identifying intellectual disabilities
McDermott, Moran [42]	**+/-**	+/- Debatable Medical records, but unclear whether it concerns a random sample or whether sample size is adequate; the matching performed was lower than 1:2	+ Sufficient Identification of intellectual disabilities via diagnosis in medical record
Perera, Audi [43]	**+/-**	+/- Debatable Use of register data, but no information on descriptive characteristics of populations	+ Sufficient Identification of intellectual disabilities via diagnosis in medical record
Dias, Ware [33]	**-**	+/- Debatable Adequacy of sample size is uncertain as there was no power calculation. There is no information on non-response, but response rate seems low	+/- Debatable Identification based on screening
Havercamp, Scandlin [38]	**-**	+/- Debatable Two different sources for groups with and without intellectual disabilities . Sample size of second group is higher than that of the first group. No information on non-response	+/- Debatable Use of three sources to identify people with intellectual disabilities, use of proxy respondents to obtain information on this group.
Morin, Mérineau-Côté [7]	**-**	+/- Debatable Random sample, but no information on non-response or response rate within study	+/- Debatable Only 7.5% of surveys were filled in by people with intellectual disabilities themselves (rest proxy). Those with intellectual disabilities not receiving any services were not included

	Identifying chronic disease	Statistical analysis
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was appropriate, but no N reported. Men with and without ID were not statistically compared, although that information was available
	+ Sufficient Chronic disease(s) identified by diagnoses in medical records	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was limited: only % was reported, no N, confidence intervals, or significance levels.
	- Insufficient Chronic disease(s) identified by self-reported values	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. The significance level was not reported
	- Insufficient Chronic disease(s) identified by self-reported values	+/- Debatable Methods were poorly described. The statistical analysis was sufficient, but the group of people with intellectual disabilities sometimes had low response
	- Insufficient Chronic disease(s) identified by self-reported values	+/- Debatable Methods were described in sufficient detail to properly identify the analytical method. The statistical analysis was limited

CHRONIC DISEASES AND COMORBIDITIES: A CROSS-SECTIONAL STUDY IN GENERAL PRACTICE

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ABSTRACT

Introduction: Chronic disease and comorbidity patterns in people with intellectual disabilities (ID) are more complex than in the general population. However, incomplete understanding of these differences limit care providers in addressing them.

Objective: To compare chronic disease and comorbidity patterns in chronically ill patients with and without ID in Dutch general practice.

Methods: In this population-based study, a multi-regional primary care database of 2018 was combined with national population data to improve identification of adults with ID. Prevalence was calculated using Poisson regression to estimate prevalence ratios and 95% confidence intervals for the highest-impact chronic diseases (ischemic heart disease (IHD), cerebrovascular disease (CVD), diabetes mellitus (DM), and chronic obstructive pulmonary disease (COPD)) and comorbidities.

Results: Information from 18,114 people with ID and 1,093,995 people without ID was available. When considering age and sex, CVD (PR=1.1), DM (PR=1.6), and COPD (PR=1.5) times more prevalent in people with than without ID. At younger age, people with ID more often had a chronic disease and multiple comorbidities. Males with ID most often had a chronic disease and multiple comorbidities. Comorbidities of circulatory nature were most common.

Conclusions: This study identified a younger onset of chronic illness and a higher prevalence of multiple comorbidities among people with ID in general practice than those without ID. This underlines the complexity of people with ID and chronic diseases in general practice. As this study confirmed the earlier onset of chronic diseases and comorbidities, it is recommended to acknowledge these age differences when following chronic disease guidelines.

INTRODUCTION

Intellectual disabilities (ID) are characterised by substantial limitations in adaptive behaviour and intellectual functioning, that are expressed in lower conceptual, social, and practical skills as compared to people without ID [1]. Approximately 1% of the global population complies to the formal definition of ID, but under-recognition and under-registration of ID could imply a higher percentage in general practice [2]. As people with ID may experience difficulties in understanding and communicating (symptoms of) diseases, it can be more challenging to diagnose and timely treat conditions, resulting in more avoidable hospitalisations and premature deaths as compared to people without ID [3-5]. Additionally, multimorbidity is highly prevalent, and frailty occurs 15 years earlier in people with ID [3, 6].

Epidemiological patterns thus substantially differ in people with versus without ID. However, a full understanding of this complexity in people with ID is hampered by incomplete and insufficient literature on several crucial aspects. The different age and sex distribution of people with ID compared with the general population, meaning their life expectancy is lower and males more often have ID than females [2, 5], should be considered when studying chronic disease patterns [7]. As current literature fails to do so, it is unclear whether the highest-impact chronic diseases, i.e. ischaemic heart disease (IHD), cerebrovascular disease (CVD), diabetes mellitus (DM), and chronic obstructive pulmonary disease (COPD), are more prevalent in people with or without ID [8, 9]. Furthermore, information on comorbidity patterns in people with ID, meaning occurrence and characteristics of additional conditions alongside a chronic disease [10], is largely lacking. Although comorbidities are highly prevalent among people with ID [11, 12] and complicate the provision of optimal healthcare, existing studies solely focus on smaller ID populations or do not consider comorbidity characteristics [12-20].

Despite these increased needs for healthcare, community-dwelling people with ID rely on the non-ID-oriented setting of regular primary care for chronic disease detection and management [21, 22]. An accurate insight in chronic diseases in people with ID is thus necessary to generate awareness among primary care providers on the need for early detection and adequately treating chronic diseases and concomitant comorbidities. This study aims to examine 1) differences in prevalence of IHD, CVD, DM, and COPD between people with and without ID, and 2) occurrences and characteristics of comorbidities in people with ID and a chronic disease compared to those without ID in Dutch primary care.

METHODS

Data sources and study population

In this cross-sectional population-based retrospective study, we selected patients registered within Nivel Primary Care Database (NPCD). Data collection took place in 2021. This database is representative of Dutch general practice by routinely collecting medical information from over a million patients registered with 420 general practices [23, 24]. Adults registered in 2018 were selected. People were identified as having ID if their medical record contained the only code available within International Classification of Primary Care (ICPC) for ID, P85 (Mental retardation) [25]. In order to improve identification of people with ID, we retrieved information on use of long-term care and supportive services from databases at Statistics Netherlands for all individuals in NPCD. If presence of an ID was noted in any of the linked databases from Statistics Netherlands, individuals were also included in the ID-group. This method is elaborated upon elsewhere [2].

Ethics

All data were pseudonymised and accessible only in Statistics Netherlands' secured research environment. This study complied with the governance orders of Nivel (NZR-00320.002) and Statistics Netherlands. Because this study concerns retrospective research with non-traceable information, Radboud university medical center's ethics committee has waived the need for formal ethical assessment (2017-3921). Prior to analyses, the research aims, hypotheses, methods, and analysis plan were preregistered (https://osf.io/kwv68/). The STROBE guidelines for reporting observational data were followed [26].

Operationalisations

Chronic diseases and comorbidities were encoded using ICPC-2 [25]. Individuals were defined as having a chronic disease when IHD, CVD, DM, and/or COPD were present in 2018 in their medical file (Supplementary Table S1). Comorbidities were defined based on a previously developed algorithm to construct illness episodes in NPCD [24], in which 109 chronic conditions were identified. ID was excluded (ICPC code P85), as it already served as selector variable, leaving 108 comorbidities (Supplementary Table S2). Comorbidity occurrence was presented as percentage of people that have 2 or more comorbidities next to their chronic disease.

Statistical analysis

Descriptive statistics of the study groups were presented as frequencies with percentages or means with standard deviations. Chronic disease prevalence was compared between people with and without ID using Poisson regression analysis, estimating prevalence

ratios (PR) and 95% confidence intervals (CIs), both unadjusted and age- and sex-adjusted. To acknowledge the large sample size, *p*-values below 0.005 were considered statistically significant [27]. The percentage of people with and without ID having a diagnosis of chronic disease was presented in percentages and shown for males and females, and in 5-year age groups. The amount of people with 2 or more comorbidities was calculated in percentages for people with and without ID for males and females, and in 5-year age groups. All analyses were conducted using SPSS (version 25.0).

RESULTS

Demographics

The study groups consisted of 18,114 people with ID and 1,093,995 people without ID (Figure 1). The percentage of males with ID (57.1%) was greater than the percentage of males without ID (48.8%) in their respective groups (Table 1). The average age of people with ID was lower than that of people without ID: 39.0 (SD: 15.9) versus 49.7 years (SD: 18.5), respectively. The majority of people with ID (71.1%) were younger than 50, with the largest group being 18–29 years (38.4%). Most of the people without ID were 50 years or older (50.4%); those aged 50–69 years were the largest group (33.9%). Of those with ID, 14.9% (n=2,653) were diagnosed with at least one chronic disease; for those without ID, this was 16.9% (n=184,681).

Table 1. Descriptive statistics of people with and without intellectual disabilities

	People with ID N=18,114	People without ID N=1,093,995
Sex, N (%)		
Males	10,336 (57.1)	534,078 (48.8)
Females	7,778 (42.9)	559,917 (51.2)
Age, N (%)		
Mean age (SD)	39.0 (15.9)	49.7 (18.5)
Age groups, N (%)		
18-49 years	12,988 (71.1)	542,620 (49.6)
18-34 years	8,911 (49.2)	276,970 (25.3)
35-49 years	4,077 (22.5)	265,650 (24.3)
50 years or older	5,126 (28.3)	551,375 (50.4)
50-69 years	4,451 (24.6)	370,442 (33.9)
70 years or older	675 (3.7)	180,933 (16.5)
People with at least one chronic disease, N (%)	2,653 (14.9)	184,681 (16.9)

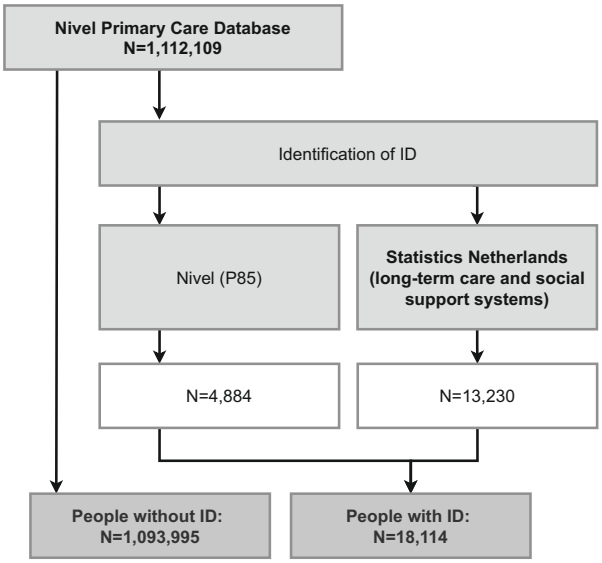

Figure 1. Identification of people with and without intellectual disabilities in final study groups

Chronic disease prevalence

Unadjusted for age and sex, IHD (PR=0.47, CI=0.43-0.51) and CVD (PR=0.69, 0.63-0.76) were less common in people with than without ID (Figure 2). People with ID more often had DM (PR=1.08, CI=1.03-1.13). There was no difference in COPD prevalence between those with and without ID (PR=0.98, CI=0.91-1.05). When adjusted for age and sex, different patterns emerged: prevalence rates all increased towards higher prevalence for people with ID. Unadjusted PRs ranged from 0.47 (IHD) to 1.08 (DM), while adjusted PRs ranged from 0.74 (IHD) to 1.62 (DM). In greater detail, CVD prevalence increased to PR=1.12 (CI=1.02-1.23), DM prevalence increased from PR=1.08 to PR=1.62 (CI=1.54-1.69), and COPD prevalence became statistically significant (PR=1.52, CI=1.42-1.63).

The onset of any of the observed chronic illnesses was at younger age for people with ID as compared to people without ID (Figure 3). In the age groups 18-24 years, prevalence was 3 to 5 times higher for people with ID than for people without ID. At ages 55-59 years, this difference was highest: to illustrate, IHD occurred in 20.2% of those with ID vs 8.3% of those without ID. Highest prevalence of any chronic illness among people with ID appeared in age groups below 70 years of age, while highest prevalence rates among people without ID are found among those aged 80 years or older. The percentage having CVD was highest among people without ID (28.3%), compared to 4.2% of those with ID (Supplementary Table S3).

Males with ID more often had a diagnosis of IHD, CVD, DM, or COPD than males without ID (not shown in figure; see Supplementary Table S3). For females with ID this pattern was reversed: females with ID less often had a diagnosis of a chronic disease than females without ID.

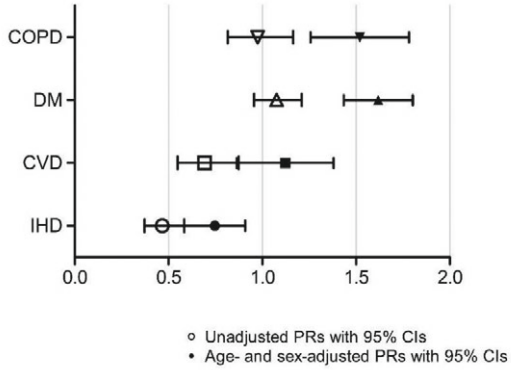

Figure 2. Chronic disease prevalence in unadjusted and sex- and age-adjusted prevalence ratios (PRs) in people with versus without intellectual disabilities

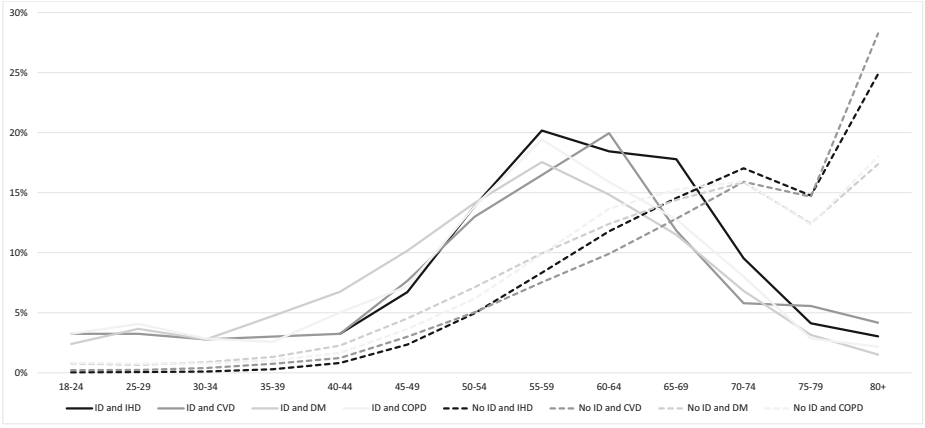

Figure 3. Percentage of people with and without intellectual disabilities that have a diagnosis of a chronic disease in 5-year age groups in 2018

Comorbidities

People with ID more often had comorbidities, at younger ages, and more often multiple comorbidities. In people with DM and COPD multiple comorbidities were more common in those with than without ID: 1.7% of 18-24-year-olds with ID and DM had 2 or more

comorbidities compared to 0.1% of those without ID. In COPD, 2.5% of 18-24-year-olds with ID had 2 or more comorbidities compared to 0.3% of people without ID (Figure 4). In people with ID, the occurrence of 2 or more comorbidities was higher before age 65, while for people without ID it was highest after age 65. The highest occurrence of 2 or more comorbidities in people with ID with DM or COPD occurred at ages 55-59 years. 17.6% of people with ID and CVD aged 60-64 years had 2 or more comorbidities. For people with ID aged 70 years or older, the percentage with 2 or more comorbidities decreased, while it increased for those without ID. Of the 80-year old people with ID and CVD, 3.5% had 2 or more comorbidities, compared to 26.5% of those without ID (Supplementary Table S4).

Males with ID more often had 2 or more comorbidities next to their chronic disease of IHD, CVD, DM, or COPD compared to those without ID (not shown in figure; see Supplementary Table S4). For females with ID this pattern was reversed: chronically ill females with ID less often had 2 or more comorbidities than those without ID.

When focusing on the characteristics of these comorbidities, comorbid diseases in the circulatory disease cluster (ICPC-code K) were most common in people with ID (not shown in figure; see Supplementary Table S4), disregarding of a diagnosis of either IHD, CVD, DM, or COPD. The most common comorbidity in all chronic diseases was hypertension, although occurrence was lower in people with than without ID. DM was also a common comorbidity in IHD, CVD, and COPD.

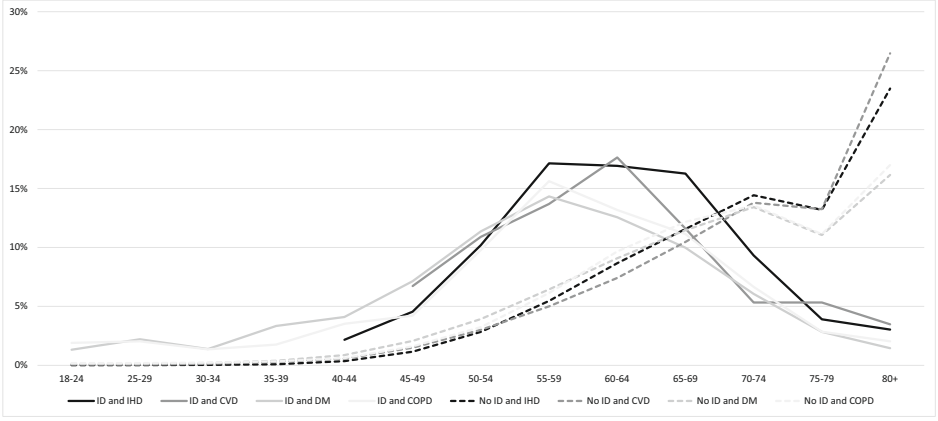

Figure 4. Percentage of people with and without intellectual disabilities having 2 or more comorbidities in 5-year age groups in 2018

DISCUSSION

Summary

This retrospective cross-sectional study examined chronic disease and comorbidity patterns in chronically ill patients with ID compared to those without ID in a Dutch general practice population in 2018. We found that although at group level chronic diseases appeared to be less prevalent in people with ID, considering age and sex revealed different patterns emerged and prevalence rates increased towards higher prevalence in those with ID. Adjusted PRs ranged from 0.74 (IHD) to 1.62 (DM). To illustrate, it seemed that already at age 18, patients with ID more often developed chronic diseases with or without comorbidities than those without ID. Although sex effects were less straightforward than age effects, results suggested that males with ID were most vulnerable: they most often had a diagnosis of chronic disease, and on top of that, they also more often had multiple comorbidities. For females, the opposite pattern emerged: the least often they were diagnosed with a chronic disease or had 2 or more comorbidities than females without ID.

Strengths and limitations

This study is the first to provide a large-scale comparative insight into comorbidity patterns in chronically ill people with and without ID in Dutch general practice. By linking a primary care database with population data, we were able to identify more individuals with ID than would be possible through their GP records alone, thereby overcoming challenges in recognising people with ID in population datasets [28]. Because of the large scale and linking possibilities, it is likely that our combined dataset provides reliable insight into clinical practice and the health of people with ID obtaining care in general practice.

Although data linkage allowed to identify more individuals with ID, the available data did not contain information on ID aetiology. This prevented us from differentiating between syndromes or ID severity, despite signs that ID severity is related to multimorbidity and specific syndromes have increased risk of specific chronic diseases [13, 21]. Future research could therefore provide a more in-depth insight by taking into account ID aetiology.

Next, this study utilised documented diagnoses to assess disease prevalence. Although these are deemed reliable [29], under-recognition of health conditions in people with ID remains a widespread concern [30]. Chronic diseases and comorbidities in people with ID may therefore not always be recognised, implying actual prevalence rates may even be higher. One way of gaining more thorough insights is to supplement data with screenings by health professionals [31], such as health assessment instruments specifically developed for people with ID to assist in diagnosing health conditions (e.g. PROSPER-ID [31]).

Comparison with existing literature

CVD, DM, and COPD were 1.5 times more prevalent in people with than without ID. These diseases, as well as IHD, are all lifestyle-related [32]. People with ID have unhealthy lifestyles more often than those without ID [33], which could possibly explain the higher DM and COPD prevalence rates. However, it does not explain the lower IHD prevalence in those with ID. It could be that cardiovascular diseases are more often managed or seen in secondary (hospital) or tertiary care settings, rather than in primary care. Some studies indeed reported higher hospitalisation for cardiovascular disease prevalence in people with ID [34, 35].

At group level chronic disease prevalence appeared to be lower in people with ID. Solely when considering age and sex, chronic disease prevalence was higher in people with than those without ID. Demographic differences between the two groups may thus influence chronic disease prevalence. How age and sex precisely affect chronic disease patterns should be further explored, though it can be the case that chronic diseases may develop at younger age in people with ID, under influence of factors such as genetics, early frailty, medicine use, or lifestyle [5, 30, 33, 36].

We confirmed previous findings on the importance of age in the prevalence of chronic diseases, such as the pattern of older people with ID more often having a chronic disease [8, 30], even at younger age. This indicates that frailty occurs earlier than in the general population [6]: at younger age, chronic diseases as well as comorbidities were more prevalent in people with ID than in the general population. At age 50-64 years, frailty occurred in similar rates in people with ID than in those without ID aged 65 years or older [6]. However, the high occurrence of COPD at young age (18-24 years) may also be due to wrongly coding asthma as COPD as it can be difficult to make the distinction between both in people with ID [37].

Having two or more comorbidities is relatively common in chronically ill people with ID [11, 13-15, 18, 19], even more so at younger age [12]. However, as most studies focus solely on older adults [13, 15, 18], comparison is difficult. This high prevalence could be associated with the congenital or genetic aetiology of the ID (i.e., epilepsy in people with ID or hypothyroidism in people with Down syndrome) [36, 38, 39].

Although our results seem to suggest that people with ID from age 65 onwards are more healthy than those without ID, a healthy survivor effect may have occurred. This means that although life expectancy of people with ID has increased, it is still lower than that of the general population [5]. The ID-population aged 65 years and older in our dataset may therefore comprise a relatively more healthy group.

While this study reported important findings regarding sex differences between those with and without ID in the prevalence of chronic diseases and comorbidities, literature is scarce. Our finding that males with ID most often had a diagnosis of a chronic disease can therefore not easily be compared to existing literature. Previous research is inconclusive on chronic conditions being more prevalent in males or females: it appears to depend on the type of conditions studied [15, 17, 19]. Additionally, unlike the current study, having two or more health conditions was found to be more common in females with ID [12, 17, 20], or others did not find a sex effect [13, 16]. Literature is thus inconclusive on sex effects in chronic diseases, however, this study highlights the importance of considering sex differences in chronic diseases between people with and without ID.

Characteristics of comorbidities in chronically ill people with ID are less often studied. We found that most comorbidities were of the circulatory system. Similar to our findings, studies reported lower prevalence of comorbidities in cardiovascular clusters in people with ID [15, 17]. As we confirmed previous literature on hypertension being a highly prevalent comorbidity in chronic diseases in people with ID [15, 17], the lower prevalence of cardiovascular comorbidities may be due to underdiagnosis.

Implications for clinical practice

People with ID display different disease patterns than the usual patients seen in general practice. Younger people with ID are particularly burdened: they more often have more chronic diseases and more comorbidities. These findings therefore aid general practitioners to develop greater awareness of differences between people with and without ID. This awareness is essential and underlying in providing suitable and tailored chronic disease management for people with ID [21, 22]. By increased collaboration between general practitioners and care professionals in specialised ID-care, recognising and treating chronic diseases and comorbidities within ID-patients can be optimised [13].

In addition, suitable chronic disease prevention and treatment could relieve the high burden of comorbidities in people with ID as presented in this study. It is therefore essential to create awareness on health behaviours and engage people with ID in lifestyle alterations to decrease body weight [33, 40]. When interventions are combined with (structural) proactive risk assessments, diseases can be diagnosed at earlier stage [31].

Conclusions

Patterns of chronic diseases with and without comorbidities were different in adults with ID compared to those without ID. It seems that people with ID developed chronic diseases with or without comorbidities at a younger age than people without ID. Males with ID carried the highest burden: they most often had a chronic disease as well as

multiple comorbidities. Due to these important differences with the general population, general practitioners should be aware of chronic diseases and comorbidities especially in younger people with ID and males with ID. Pro-active health assessments can therefore be used to timely recognise health conditions. This could be an important addition to regular chronic disease guidelines to acknowledge the earlier onset of chronic diseases and comorbidities in people with ID, and ensure their equal chances to high-quality care. This way, person-centred care can be provided, ultimately the basis for reducing existing health inequities.

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APPENDIX

Supplementary Table S1. Selection of four chronic diseases and accompanying ICPC codes

Chronic disease	ICPC code
Ischaemic heart disease	
Angina pectoris	K74
Myocardial infarction	K75
Other/chronic ischemic heart disease	K76
Cerebrovascular disease	
Transient ischemic attack	K89
Cerebrovascular accident	K90
Diabetes mellitus	T90
Chronic obstructive pulmonary disease	
Chronic bronchitis/bronchiectasis	R91
Emphysema/COPD	R95

Supplementary Table S2. Selection of 108 chronic comorbidities and accompanying ICPC codes

ICPC code	Comorbidity	ICPC code	Comorbidity
A28	Limited function/disability NOS	N28	Limited function/disability neurological
A79	Malignancy not otherwise specified	N70	Poliomyelitis
A90	Congenital anomaly otherwise specified/multiple	N74	Malignant neoplasm nervous system
B28	Limited function/disability blood/ lymphatic system	N85	Congenital anomaly neurological
B72	Hodgkin disease/lymphoma	N86	Multiple sclerosis
B73	Leukaemia	N87	Parkinsonism
B74	Malignant neoplasm blood other	N88	Epilepsy
B78	Hereditary haemolytic anaemia	P28	Limited function/disability psychological
B79	Congenital anomaly blood/lymphatic system other	P70	Dementia
B83	Purpura/coagulation defect	P72	Schizophrenia
B90	HIV-infection/aids	P80	Personality disorder
D28	Limited function/disability digestive	R28	Limited function/disability respiratory system
D74	Malignant neoplasm stomach	R84	Malignant neoplasm bronchus/lung
D75	Malignant neoplasm colon/rectum	R85	Malignant neoplasm respiratory, other
D76	Malignant neoplasm pancreas	R89	Congenital anomaly respiratory tract
D77	Malignant neoplasm digestive system other/not otherwise specified	R91	Chronic bronchitis/bronchiectasis
D81	Congenital anomaly digestive system	R95	Chronic obstructive pulmonary disease
D92	Diverticular disease	R96	Asthma
D94	Chronic enteritis/ulcerative colitis	S28	Limited function/disability skin
D97	Liver disease not otherwise specified	S77	Malignant neoplasm of skin
F28	Limited function/disability eye	S81	Haemangioma/lymphangioma
F81	Congenital abnormality eye/other	S83	Congenital skin anomaly other
F83	Retinopathy	S87	Dermatitis/atopic eczema
F84	Macular degeneration	S91	Psoriasis
F91	Refractive error	T28	Limited function/disability endocrine system/metabolism/nutrition
F93	Glaucoma	T71	Malignant neoplasm thyroid
F94	Blindness	T78	Thyroglossal duct/cyst

Supplementary Table S2. Continued

ICPC code	Comorbidity	ICPC code	Comorbidity
H28	Limited function/disability ear	T80	Congenital anomaly endocrine/ metabolic system
H80	Congenital anomaly of ear	T81	Goitre
H83	Otosclerosis	T86	Hypothyroidism/myxoedema
H84	Presbycusis	T90	Diabetes mellitus
H85	Acoustic trauma	T92	Gout
H86	Deafness	T93	Lipid disorder
K28	Limited function/disability cardiovascular	U28	Limited function/disability urinary tracts
K73	Congenital anomaly cardiovascular system	U75	Malignant neoplasm of kidney
K74	Ischaemic heart disease with angina pectoris	U76	Malignant neoplasm of bladder
K76	Ischaemic heart disease without angina pectoris	U77	Malignant neoplasm urinary other
K77	Heart failure	U85	Congenital anomaly urinary tracts
K82	Pulmonary heart disease	U88	Glomerulonephritis/nephrosis
K86	Hypertension uncomplicated	W28	Limited function/disability as a result of pregnancy
K87	Hypertension complicated	W72	Malignant neoplasm related to pregnancy
K90	Stroke/cerebrovascular accident	W76	Congenital anomaly complicating pregnancy
K91	Atherosclerosis/PVD	X28	Limited function/disability female genitals
K92	Pulmonary embolism	X75	Malignant neoplasm cervix
L28	Limited function/disability musculoskeletal	X76	Malignant neoplasm breast female
L82	Congenital anomaly musculoskeletal system	X77	Malignant neoplasm genital other (f)
L84	Back syndrome without radiating pain	X83	Congenital anomaly genital female
L85	Acquired deformity of spine	X88	Fibrocystic disease breast
L88	Rheumatoid/seropositive arthritis	Y28	Limited function/disability male genitals
L89	Osteoarthrosis of hip	Y77	Malignant neoplasm of prostate
L90	Osteoarthrosis of knee	Y78	Malignant neoplasm male genital other

Supplementary Table S2. Continued

ICPC code	Comorbidity	ICPC code	Comorbidity
L91	Osteoarthrosis other	Y82	Hypospadias
L95	Osteoporosis	Y84	Congenital genital anomaly other (m)
L98	Acquired deformity of limb	Z28	Limited function/disability social problems

Table S3. Prevalence of chronic diseases for people with and without intellectual disabilities by sex and 5-year age groups in N (%)

	IHD	
	ID	No ID
	N=18,114	N=1,093,995
Unadjusted PR (95% CIs)	**0.466** (0.425; 0.510)**	
Adjusted PR (95% CIs)	**0.744** (0.681; 0.812)**	
Total group	461 (2.5)	59,808 (5.5)
Sex		
Males	325 (3.1)	37,407 (7.0)
Females	136 (1.7)	22,401 (4.0)
Age groups		
18-24 years	<10	26 (<0.1)
25-29 years	<10	39 (0.1)
30-34 years	<10	65 (0.1)
35-39 years	<10	181 (0.3)
40-44 years	15 (3.3)	494 (0.8)
45-49 years	31 (6.7)	1406 (2.4)
50-54 years	64 (13.9)	2965 (5.0)
55-59 years	93 (20.2)	4984 (8.3)
60-64 years	85 (18.4)	7050 (11.8)
65-69 years	82 (17.8)	8708 (14.6)
70-74 years	44 (9.5)	10184 (17.0)
75-79 years	19 (4.1)	8827 (14.8)
80+ years	14 (3.0)	14879 (24.9)

CVD		DM		COPD	
ID	No ID	ID	No ID	ID	No ID
N=18,114	N=1,093,995	N=18,114	N=1,093,995	N=18,114	N=1,093,995
0.692 (0.630; 0.760)**		**1.076** (1.026; 1.128)**		0.975 (0.907; 1.047)	
1.118* (1.020; 1.227)		**1.616** (1.544; 1.692)**		**1.517** (1.415; 1.626)**	
431 (2.4)	37,613 (3.4)	1,584 (8.7)	88,925 (8.1)	736 (4.1)	45,611 (4.2)
255 (2.5)	18,978 (3.6)	874 (8.5)	47,125 (8.8)	428 (4.1)	22,713 (4.3)
176 (2.3)	18,635 (3.3)	710 (9.1)	41,800 (7.5)	308 (4.0)	22,898 (4.1)
14 (3.2)	84 (0.2)	38 (2.4)	694 (0.8)	24 (3.3)	383 (0.8)
14 (3.2)	91 (0.2)	58 (3.7)	588 (0.7)	30 (4.1)	344 (0.8)
12 (2.8)	149 (0.4)	44 (2.8)	786 (0.9)	21 (2.9)	361 (0.8)
13 (3.0)	283 (0.8)	75 (4.7)	1176 (1.3)	19 (2.6)	472 (1.0)
14 (3.2)	464 (1.2)	107 (6.8)	2024 (2.3)	37 (5.0)	756 (1.7)
33 (7.7)	1129 (3.0)	161 (10.2)	4023 (4.5)	53 (7.2)	1661 (3.6)
56 (13.0)	1895 (5.0)	224 (14.1)	6336 (7.1)	102 (13.9)	2817 (6.2)
71 (16.5)	2835 (7.5)	278 (17.6)	8847 (9.9)	143 (19.4)	4502 (9.9)
86 (20.0)	3727 (9.9)	235 (14.8)	11025 (12.4)	117 (15.9)	6235 (13.7)
51 (11.8)	4827 (12.8)	182 (11.5)	12803 (14.4)	94 (12.8)	6961 (15.3)
25 (5.8)	5984 (15.9)	108 (6.8)	14113 (15.9)	59 (8.0)	7268 (15.9)
24 (5.6)	5515 (14.7)	50 (3.2)	11058 (12.4)	21 (2.9)	5635 (12.4)
18 (4.2)	10630 (28.3)	24 (1.5)	15452 (17.4)	16 (2.2)	8216 (18.0)

Table S4. Amount of chronically ill people with and without intellectual disabilities having 2 or more comorbidities and comorbidity characteristics by sex and 5-year age groups in N (%)

	IHD	
	ID	No ID
People having 2+ comorbidities		
Total group	395 (85.7)	48645 (81.3)
Sex		
Males	270 (68.4)	29021 (59.7)
Females	125 (31.6)	19624 (40.3)
Age groups		
18-24 years	<10	10 (<0.1)
25-29 years	<10	13 (<0.1)
30-34 years	<10	24 (<0.1)
35-39 years	<10	57 (0.1)
40-44 years	10 (2.5)	214 (0.4)
45-49 years	21 (5.3)	693 (1.4)
50-54 years	47 (11.9)	1696 (3.5)
55-59 years	79 (20.0)	3267 (6.7)
60-64 years	78 (19.7)	5183 (10.7)
65-69 years	75 (19.0)	6921 (14.2)
70-74 years	43 (10.9)	8632 (17.7)
75-79 years	18 (4.6)	7891 (16.2)
80+ years	14 (3.5)	14044 (28.9)
Comorbidity characteristics		
Circulatory diseases	279 (60.5)	39,484 (66.0)
Hypertension	170 (36.9)	25,028 (41.8)
DM	165 (35.8)	16,611 (27.8)

	CVD		DM		COPD	
	ID	No ID	ID	No ID	ID	No ID
	363 (84.2)	30865 (82.1)	1237 (78.1)	66995 (75.3)	560 (76.1)	34688 (76.1)
	204 (56.2)	15118 (49.0)	645 (52.1)	33872 (50.6)	317 (56.6)	16808 (48.5)
	159 (43.8)	15747 (51.0)	592 (47.9)	33123 (49.4)	243 (43.4)	17880 (51.5)
	<10	26 (0.1)	21 (1.7)	101 (0.2)	14 (2.5)	89 (0.3)
	<10	22 (0.1)	35 (2.8)	116 (0.2)	15 (2.7)	87 (0.3)
	<10	48 (0.2)	22 (1.8)	180 (0.3)	10 (1.8)	105 (0.3)
	<10	115 (0.4)	53 (4.3)	343 (0.5)	13 (2.3)	152 (0.4)
	<10	211 (0.7)	65 (5.3)	775 (1.2)	26 (4.6)	278 (0.8)
	29 (8.0)	562 (1.8)	113 (9.1)	1844 (2.8)	31 (5.5)	735 (2.1)
	47 (12.9)	1135 (3.7)	180 (14.6)	3493 (5.2)	72 (12.9)	1477 (4.3)
	59 (16.3)	1884 (6.1)	227 (18.4)	5728 (8.5)	115 (20.5)	2798 (8.1)
	76 (20.9)	2790 (9.0)	199 (16.1)	8080 (12.1)	97 (17.3)	4406 (12.7)
	50 (13.8)	3940 (12.8)	158 (12.8)	10199 (15.2)	82 (11.1)	5552 (12.2)
	23 (6.3)	5190 (16.8)	96 (7.8)	11942 (17.8)	49 (6.7)	6182 (13.6)
	23 (6.3)	4986 (16.2)	45 (3.6)	9830 (14.7)	21 (2.9)	5074 (11.1)
	15 (4.1)	9956 (32.3)	23 (1.9)	14364 (21.4)	15 (2.0)	7753 (17.0)
	247 (57.3)	26,431 (70.3)	812 (51.3)	60,645 (68.2)	350 (47.6)	27,558 (60.4)
	142 (32.9)	16,563 (44.0)	568 (35.9)	44,147 (49.6)	211 (28.7)	17,529 (38.4)
	102 (23.7)	8,861 (23.6)			166 (22.6)	9,137 (20.0)

PART II

CHRONIC DISEASE MANAGEMENT

RECORDS OF QUALITY INDICATORS: DISEASE MONITORING IN RESIDENTIAL CARE

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ABSTRACT

Introduction: Type 2 diabetes mellitus, cardiovascular disease, and chronic obstructive pulmonary disease (COPD) contribute significantly to societal and individual impact globally. High-quality management of these long-term health conditions is important to prevent deterioration of health, although potentially more complex for patients with intellectual disabilities in residential care. Disease management in this context particularly benefits from complete and accurate recording of disease management. Without complete records, long-term health conditions are more difficult to track due to the level of uncertainty regarding which clinical examinations have and have not been performed. This study therefore aims to examine the recording routines of quality indicators for disease monitoring for chronically ill patients with intellectual disabilities in Dutch residential care.

Methods: This retrospective study utilised medical record data from a large Dutch long-term care provider. We assessed the occurrence of cardiovascular disease (ICPC-2 codes K74, K75, K76, K89, K90), type 2 diabetes mellitus (T90, T90.02), and/or COPD (R91, R95). For adults with intellectual disabilities and a long-term condition, we analysed data entries in an 18-month period (between July 2020 and December 2021). Observed consultation rates were calculated and presented in median with interquartile range (IQR), and contrasted against the baseline number of consultation in primary care. Information on recorded quality indicators was presented in frequencies and percentages.

Findings: Of the three long-term conditions investigated, the most common was type 2 diabetes mellitus (8.6%; n=287), followed by cardiovascular disease (5.8%; n=195), and COPD (3.0%; n=101). Of those who received management for their long-term condition from their contracted GP, patients with type 2 diabetes mellitus, cardiovascular disease, or COPD had fewer consultations in 2021 than the Dutch baseline. Discussion of lifestyle was often not recorded. Disease monitoring quality indicators were recorded more often, but at a lower frequency than expected.

Conclusions: Because of the infrequent recording of quality indicators, recording of management of long-term conditions for patients with intellectual disabilities in long-term care appears suboptimal. Although this may not directly harm individual patients, it may jeopardise the quality of management of long-term conditions, as suboptimal recording limits opportunities for evaluation and improvement. Within a broader trend towards data-driven work methods in healthcare, recording of quality indicators requires attention from practice, research, and policy.

INTRODUCTION

The impact of long-term health conditions globally is high, especially type 2 diabetes mellitus, cardiovascular disease, and chronic obstructive pulmonary disease (COPD). They are among the most prevalent long-term health conditions globally, with prevalence rates expecting to increase [1]. Collectively these three health conditions account for 22 million deaths per year [2-4], and contribute to high healthcare expenditures [5]. They may also decrease patients' mental health, social functioning and health-related quality of life [6, 7].

For patients with intellectual disabilities, the impact of these long-term health conditions may even be higher than for the general population. Their limitations in intellectual functioning and adaptive behaviour [8] may partly contribute to increased risk for long-term health conditions, increased prevalence of health problems, and increased care needs [9-12]. Unhealthy lifestyle, the main risk factor for development of these health conditions, is even more common among people with than without intellectual disabilities [13-15]. About 1-3% of the global population has a diagnosis of intellectual disabilities; in the Dutch population around 1.5% [16, 17]. Almost half of all people with a diagnosis of intellectual disabilities in the Netherlands receives care in residential care settings [17]. Those people are often characterised by more severe intellectual disabilities and increased healthcare needs, amplifying their need for adequate disease monitoring [10, 18, 19]. To reduce this impact, it is essential that management of these health conditions is of high quality for all patients, especially those with intellectual disabilities. In the Netherlands, these long-term health conditions are being managed in general practice, in which standardised disease management programmes sustain the high quality of care [20, 21]. Disease management programs provide patients with continuous, patient-centred, and comprehensive care, in which quality is assured by the use of quality indicators [21]. Similar to the Quality and Outcomes Framework in the UK, these indicators are developed to ensure quality of management of long-term conditions by supporting healthcare professionals in compliance to evidence-based guidelines, by assessing technical aspects of care (such as use of suitable medication), and by supporting patients in attaining most optimal disease control [22, 23]. Through financial incentives healthcare professionals are stimulated to regularly monitor long-term health conditions through these indicators, and to record the performance and results of these tests in patient's medical records.

However, for patients with intellectual disabilities, the quality of care can be contested as inequalities in the quality of management of these long-term health conditions still exist between people with and without intellectual disabilities. Studies reported less checking of cholesterol and blood pressure in those with cardiovascular disease and intellectual

disabilities [24, 25], fewer spirometry tests and checking of inhaler technique in those with COPD and intellectual disabilities [24, 25], and fewer tests of HbA1c, albuminuria, or eye exams in those with type 2 diabetes mellitus and intellectual disabilities [24-28]. Lifestyle is also not often discussed in people with intellectual disabilities. For example, they were less likely to receive smoking cessation advice [24, 25], and physical activity levels and healthy dietary patterns continue to remain lower than of those without intellectual disabilities [14, 15]. Cardiovascular risk factors remain undiagnosed almost twice as often in the population with intellectual disabilities compared to the general population [29], probably due to people with intellectual disabilities receiving fewer preventative medical screenings [30-32].

These lower frequencies of performed clinical examinations are disadvantageous for the already worse health of people with intellectual disabilities. Although the same guidelines for health conditions as in regular primary care apply in residential care settings, the complexity of patients with intellectual disabilities in residential care can cause healthcare professionals to feel less equipped to deliver high quality care to these patients [33]. Additionally, systematic quality evaluation is lacking due to the absence of disease management programs and concomitant incentives to record quality indicators in patients' medical files. This leaves clinical practice unable to ensure quality of the management of long-term health conditions if recordings of quality indicators are incomplete. Inaccurate records may result in performing the same clinical examinations multiple times, thereby unnecessarily taxing the patients as well as generating high healthcare costs.

By exploring the recording of quality indicators, this study lays the foundations for suitable management of long-term conditions for patients with intellectual disabilities. To the best of our knowledge, no studies have yet shed light on monitoring patterns for long-term health conditions in people with intellectual disabilities in residential care settings. This study therefore aims to examine the current state of affairs in recordings of management of long-term conditions in long-term care settings for people with intellectual disabilities.

METHODS

Data source and participants

This retrospective study utilised patient-level medical record data from 's Heeren Loo, one of the largest long-term care providers for people with intellectual disabilities in the Netherlands. At this residential care organisation either individual general practitioners or general practices were contracted to provide on-site primary medical care, including

management of long-term conditions. Although general practitioners carried the main responsibility for managing health conditions, tasks such as disease monitoring, signalling complications, and patient education could be delegated to practice nurses [34]. From the electronic medical record system Promedico, selected pseudonymised data originating from July 2020 to December 2021 was extracted. This study complied to STROBE guidelines for cross-sectional studies [35].

People aged 18 years or older, diagnosed with a health condition, living on site at 's Heeren Loo because of an intellectual disability, and enrolled in Promedico (i.e., their GP providing care for their health condition) were selected (Figure 1). Prior to commencement of this study, the scientific board of 's Heeren Loo approved the study protocol and did not report any ethical issues. Due to the retrospective nature of this study and the use of non-identifiable information, the Radboud University Medical Centre's ethics committee waived the need for formal ethical assessment (2017-3921).

Measurements

Prevalence of long-term health conditions. In the total study population, we examined the occurrence of three long-term health conditions. Diagnoses included cardiovascular disease (International Classification for Primary Care 2 (ICPC-2 [36]) codes K74, K75, K76, K89, K90)), type 2 diabetes mellitus (T90, T90.02), and/or COPD (R91, R95).

Consultation rates. Within the group of patients with a diagnosis of a health condition, consultation rates were examined, defined as (night) consults, e-consults, (night) telephone consults, and (night) visits by a GP. The observed number of consultations was calculated per patient per year for patients with a long-term condition who had at least one consultation between July 2020 and December 2021. We displayed the median number of consultations per year. Consultation rates were weighted by registration time by dividing the number of observed consultations per patient by their registration time in the medical record system. This allowed us to also include patients with a registration time of less than 18 months. We displayed observed consultations against the baseline number of consultations in primary care for patients with a long-term health condition: nine consultations for patients with type 2 diabetes mellitus, eight for patients with cardiovascular disease, and seven for patients with COPD [37, 38].

Recording of quality indicators. Within the group of patients with a diagnosis of a long-term condition, we examined the frequency of recorded process quality indicators in patients' medical records. We distinguished between quality indicators related to disease monitoring and related to lifestyle. We selected those that were advised to be monitored in patients with a long-term condition, according to Dutch primary

care guidelines that were practiced in 2021 [39-41]. These are considered essential for evaluating the quality of chronic disease management in regular general practice [42]. A list of codes for quality indicators can be found in Appendix Table A1. The quality indicators for monitoring of cardiovascular disease included a record of a blood pressure, eGFR, albuminuria, and glucose test. The indicators for monitoring of type 2 diabetes mellitus included a record of test for blood pressure, eGFR, albuminuria, HbA1c, foot examination, and fundus exam. For monitoring of COPD, the indicators included the record of disease burden, record of daily functioning, number of exacerbations, and spirometry. The quality indicators related to lifestyle included a discussion of smoking behaviour, dietary pattern, physical activity, alcohol use, and a BMI test. As guidelines for long-term conditions did not systematically include all of the above-mentioned lifestyle indicators for all long-term health conditions, we included only those specifically mentioned in the separate cardiovascular disease, type 2 diabetes mellitus, or COPD guidelines. Recording of quality indicators was displayed for patients with a health condition that had at least one consultation between July 2020 and December 2021, to ensure that patients did not have an external healthcare provider monitoring their disease (i.e., a healthcare provider not employed at the residential care organisation). This allowed us more robustly to make claims on recording of quality indicators, as it can be expected that those with a long-term health conditions visit their GP at least once in 18 months.

Analyses

Descriptive statistics of the study groups (sex, age, and number of people having at least one consultation) were presented in frequencies and percentages. Information on the recording of consultation rates and recorded quality indicators were presented only for patients who had a diagnosis of cardiovascular disease, type 2 diabetes mellitus, or COPD, and who had a record of at least one consultation in the observed time period. Stratified by diagnosis, the median observed number of actual consultations per year was displayed, next to the baseline number of consultations per year. Interquartile range (IQR) was displayed to illustrate the range in the observed consultation rates. Stratified by diagnosis, quality indicators were presented in frequencies and percentages. All analyses were performed in SPSS version 25.0.

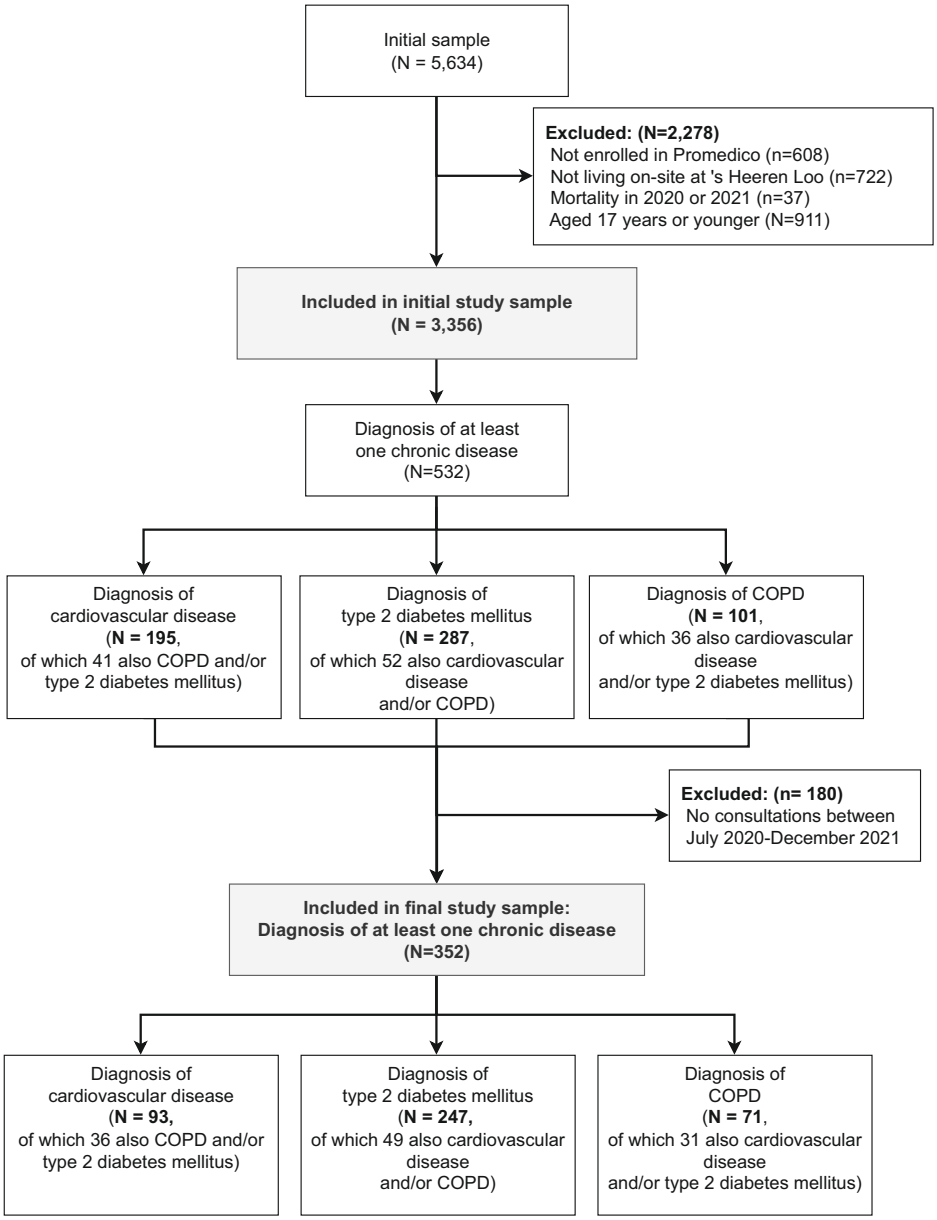

Figure 1. Flow chart of initial to final sample of study participants

RESULTS

Prevalence of long-term conditions

Of the total study population (n=3,356), most were male (56.7%; n=1,902) and were between 55-74 years old (32.8%; n=1,100; Table 1). In the total study population, 352 people had a diagnosis of at least one health condition. Of the three long-term conditions studied, most had a diagnosis of type 2 diabetes mellitus (8.6%; n=287), followed by cardiovascular disease (5.8%; n=195) and COPD (3.0%; n=101). More males than females had cardiovascular disease (54.4% vs. 45.6%) or COPD (53.5% vs. 46.5%). More females than males had a diagnosis of type 2 diabetes mellitus (58.5% vs. 41.5%). Patients aged 55-74 most often had a diagnosis of cardiovascular disease (61.0%; n=119), diabetes (48.1%; n=138) or COPD (56.4%; n=57). Most were registered for the entire study period of 18 months; 97.6% of patients with type 2 diabetes mellitus (n=241), 97.2% of patients with COPD (n=69), and 93.5% of patients with cardiovascular disease (n=87).

Table 1. Descriptive statistics and prevalence of long-term health conditions in people with intellectual disabilities living in residential care facilities in N (%)

	Total study population N=3,356	Cardiovascular disease N=195	Type 2 diabetes mellitus N=287	COPD N=101
Sex				
Males	1,902 (56.7)	106 (54.4)	119 (41.5)	54 (53.5)
Females	1,454 (43.3)	89 (45.6)	168 (58.5)	47 (46.5)
Age				
18-34 years	1,096 (32.7)	11 (5.6)	30 (10.5)	<10
35-54 years	1,024 (30.5)	41 (21.0)	99 (34.5)	28 (27.7)
55-74 years	1,100 (32.8)	119 (61.0)	138 (48.1)	57 (56.4)
75 years or older	136 (4.1)	24 (12.3)	20 (7.0)	12 (11.9)
At least 1 consultation		93 (47.7)	247 (86.1)	71 (70.3)
Of whom ≥18 months registered at GP		87 (93.5)	241 (97.6)	69 (97.2)
Disease prevalence				
Cardiovascular disease	195 (5.8)			
Type 2 diabetes mellitus	287 (8.6)			
COPD	101 (3.0)			

GP=general practice; COPD = chronic obstructive pulmonary disease.

Records of consultation rates

Patients with cardiovascular disease were recorded to have had a median of 2.0 consultations (IQR=1.3-6.8) in 2021 (Figure 2). The baseline number of consultations per patient with cardiovascular disease was eight consultations in 2021. Among patients with type 2 diabetes mellitus we observed a median of 4.7 consultations (IQR=2.7-8.7), compared to the baseline of nine consultations per year. Patients with COPD were recorded to have had a median of 2.7 consultations (IQR=1.3-9.3), while for individual patients the baseline number was seven consultations.

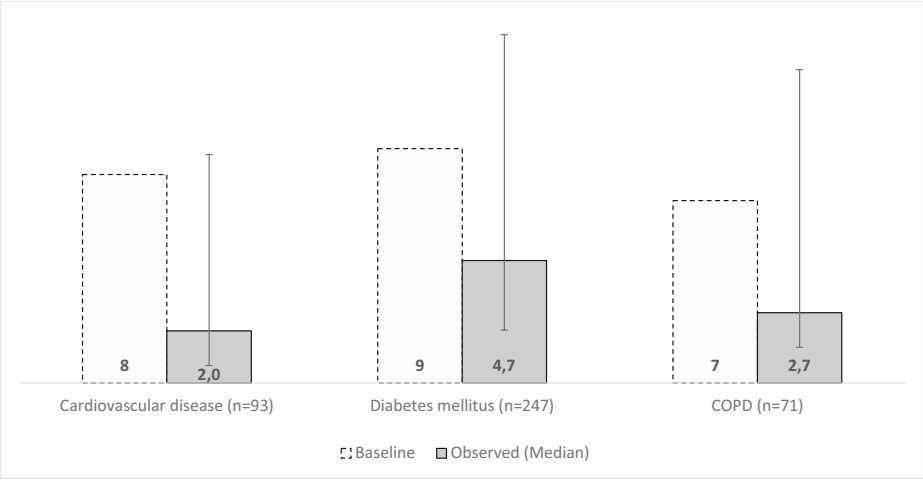

Figure 2. Median number of recorded consultations and interquartile range in 2021† of people with intellectual disabilities and a long-term health condition living in residential care facilities, compared with the baseline number of consultations in 2021‡

† Rates were weighted by registration time to show frequency per year, by dividing the number of observed consultations per patient by their registration time in the medical record system. Consultation rates were calculated for patients with at least one consultation over the 18-month period between July 2020 and December 2021.

‡ The baseline number of consultations in primary care for patients with a long-term health condition was nine consultations for patients with type 2 diabetes mellitus, eight for patients with cardiovascular disease, and seven for patients with COPD [37, 38].

Quality indicators

Table 2 displays the recording of quality indicators as recommended in the Dutch 2021 guidelines for long-term conditions. A complete recorded profile of all advised quality indicators for disease monitoring and lifestyle was recorded for only a few patients with cardiovascular disease (n=<10) or type 2 diabetes mellitus (n=<10), and none with COPD.

In 9.3% (n=34) of patients with cardiovascular disease all disease-specific quality indicators were recorded. Glucose (96.8%; n=90), eGFR (95.7%; n=89), and blood pressure tests (79.6%; n=74) were most often recorded. While BMI test was recorded for 57.0% of cardiovascular patients (n=53), other lifestyle discussions were less often recorded (smoking behaviour 49.5% (n=46); alcohol use 32.3% (n=30); dietary pattern and physical activity discussed in less than 10 patients).

In 7.1% (n=26) of patients with type 2 diabetes mellitus, all diabetes-specific quality indicators were recorded. eGFR (90.7%; n=224), HbA1c (86.6%; n=214), and blood pressure (80.2%; n=198) were most often recorded. Respectively 14.6% (n=36) and 21.5% (n=53) of diabetes patients were recorded to have had received a foot or fundus exam. Of all lifestyle quality indicators, BMI test (74.9%, n=185), discussion of smoking behaviour (49.4%; n=122), and of alcohol use (32.4%; n=80) were most often recorded.

For no patients with COPD was a complete profile of quality indicators recorded. Smoking behaviour was recorded in 52.1% (n=37), and 45.1% of patients had a record of a BMI test (n=32). No patients with COPD had a record of disease burden, exacerbations, or spirometry.

Table 2. Recorded quality indicators for disease monitoring between July 2020 and December 2021in chronically ill patients with intellectual disabilities living in residential care facilities (who had at least one consultation) as recommended in the Dutch 2021 guidelines for long-term conditions in N (%)†

	N (%)
Cardiovascular disease	N=93
Patients with complete profile of lifestyle and monitoring indicators (smoking behaviour, diet, physical activity, alcohol use, BMI, blood pressure, glucose, eGFR, albuminuria)	*<10*
Patients with complete profile of cardiovascular disease specific indicators (blood pressure, glucose, eGFR, albuminuria)	*34 (9.3)*
Smoking behaviour ever discussed	46 (49.5)
Dietary pattern discussed in past 12 months	<10
Physical activity discussed in past 12 months	<10
Alcohol use discussed in past 5 years (2017-2021)	30 (32.3)
BMI test in past 12 months	53 (57.0)
Blood pressure test in past 12 months	74 (79.6)
eGFR test in past 5 years (2017-2021)	89 (95.7)
Albuminuria test (albumin/creatinine ratio) in past 12 months	38 (40.9)
Glucose test in past 5 years (2017-2021)	90 (96.8)

Table 2. Continued

	N (%)
Cardiovascular disease	N=93
Type 2 diabetes mellitus	N=247
Patients with complete profile of lifestyle and monitoring indicators (smoking behaviour, diet, physical activity, alcohol use, blood pressure, eGFR, albuminuria, HbA1c, foot examination, fundus exam)	*<10*
Patients with complete profile of diabetes specific indicators (blood pressure, eGFR, albuminuria, HbA1c, foot examination, fundus exam)	*26 (7.1)*
Smoking behaviour ever discussed	122 (49.4)
Dietary pattern discussed in past 12 months	<10
Physical activity discussed in past 12 months	44 (17.8)
Alcohol use discussed in past 5 years (2017-2021)	80 (32.4)
BMI test in past 12 months	185 (74.9)
Blood pressure test in past 12 months	198 (80.2)
eGFR test in past 12 months	224 (90.7)
Albuminuria test (albumin/creatinine ratio) in past 12 months	153 (61.9)
HbA1c test in past 12 months	214 (86.6)
Foot examination in past 12 months	36 (14.6)
Fundus exam in past 36 months (2019-2021)	53 (21.5)
COPD	N=71
Patients with complete profile of lifestyle and monitoring indicators (smoking behaviour, physical activity, BMI, disease burden, daily functioning, exacerbations, spirometry)	*0 (0.0)*
Patients with complete profile of COPD specific indicators (disease burden, daily functioning, exacerbations, spirometry)	*<10*
Smoking behaviour ever discussed	37 (52.1)
Physical activity discussed in past 12 months	<10
BMI test in past 12 months	32 (45.1)
Record of disease burden in past 12 months	0 (0.0)
Record of daily functioning in past 12 months (CCQ or MRC)	<10
Number of exacerbations recorded in past 12 months	0 (0.0)
Spirometry in past 36 months (2019-2021)	0 (0.0)

[†] Selected quality indicators for disease monitoring were based on 2021 guidelines for cardiovascular disease (CVRM), type 2 diabetes mellitus, and chronic obstructive pulmonary disease (COPD) by Dutch College of General Practitioners [39-41]. Records of selected indicators was calculated over the 18-month period between July 2020 and December 2021 for patients with at least one consultation.

DISCUSSION

This descriptive study examined the recording of quality indicators for management of cardiovascular disease, type 2 diabetes mellitus, and COPD in patients with intellectual disabilities in residential care facilities. We reported at least four important findings related to access to care and recording of quality indicators.

First, related to access to care, the observed median number of consultations for patients with a long-term health condition was relatively low considering the baseline. As the baseline number of consultations was based on the general population in routine primary care, and not on patients with intellectual disabilities, this comparison may not be fully suitable in residential care. Previous research is inconclusive about the actual number of consultations in primary care among people with intellectual disabilities: rates ranged from an average of 1.9 to 23.8 consultations per year [43, 44]. Although patients with intellectual disabilities generally appear to have higher consultation rates than patients without intellectual disabilities [44-46], future research is invited to investigate the extent to which these discrepancies between consultation rates impact quality of care. Also, potentially incomplete recording of consultations might reflect a skewed image of reality. Recently, researchers observed a trend in the Netherlands towards less recording of management of long-term conditions, using the same baseline of consultations as we did [37]. However, the reason for this trend is yet unknown.

Second, we found that the overall recording of quality indicators was often incomplete. On the one hand, this could indicate that disease monitoring is less structured in people with intellectual disabilities than for the general population. Difficulties in performing clinical examinations in people with intellectual disabilities [47-50], often results in GPs having to choose which aspects of disease management are most important to tackle short-term, rather than establishing more long-term goals. On the other hand, information systems may be too elaborate for care for people with intellectual disabilities, and they often do not allow for reasonable adjustments [51]. Healthcare professionals can be discouraged from appropriately registering all quality indicators due to issues such as high administrative burden in care for people with intellectual disabilities [52], low experienced user friendliness of health information systems, such as the one used in this study [53], and difficulties in collaboration across care organisations due to the use of different electronic patient records [54]. More comprehensive recording of quality indicators would allow healthcare professionals to more effectively prevent comorbidities and complications.

It is difficult to compare our findings to existing studies on disease monitoring, since this is not often studied in people with intellectual disabilities. Studies that did examine disease monitoring among this patient population often reported highly fluctuating rates [24, 28]. Existing literature seems to indicate worse compliance with guidelines for people with than without intellectual disabilities: monitoring for diabetes was less often recorded than in the general population [24-28], as was monitoring for COPD [24, 25]. This is in line with our finding that recording of quality indicators for COPD monitoring was most often incomplete. It may be the case that COPD monitoring indicators as advocated in the guidelines, such as spirometry or questionnaires on disease burden, may not be suitable for people with intellectual disabilities.

Third, test results from laboratories provided useful insights into received management of long-term conditions. Outcomes of blood or urine tests (eGFR, HbA1c, and albuminuria) were automatically added to patients' medical records, providing a more accurate picture regarding the performance of those particular tests. We found a similar rate of patients with type 2 diabetes mellitus having received an HbA1c test (76.2%) in other studies using administrative data sources [24, 26]. Comparative data for other DM indicators is scarce (i.e. eGFR, albuminuria test) [24, 26, 27]. It is at least worth noting that we found that patients with type 2 diabetes mellitus in our study more often received albuminuria tests than those with cardiovascular disease did, despite guidelines for both health conditions suggesting regular albuminuria tests, and thus expecting similar rates in both disease groups. Future research is invited to shed additional light on these inconsistencies.

Fourth, records of discussion of (healthy) lifestyle between patients and healthcare professionals were often missing. The relatively unhealthy lifestyle of people with intellectual disabilities is widely documented: they are more often overweight or obese, more often smoke, and are more often physically inactive [e.g. 13, 14, 15]. Promoting healthy lifestyles is thus of high importance, as maintaining a healthy lifestyle functions as an important but often underestimated instrument in effective disease management [55-57]. Although such conversations between patients with intellectual disabilities and healthcare professionals have been reported to take place only seldom [24], it may be the case that they are either simply not recorded, are recorded outside the officially designated location within patients' medical records, or took place as part of another consultation (e.g. blood pressure measurement) but not recorded separately. Discussing lifestyle, such as smoking behaviour, may also be more difficult in people with intellectual disabilities, as they may have difficulties in understanding the relation between health and smoking [58]. Additionally, for people with more severe intellectual disabilities, the restrictiveness of their living environments (in terms of rigid procedures for staff support and activity

planning) seems to be more important in healthy lifestyle rather than discussing lifestyle [59]. Contextual factors, such as strict smoking policies, may leave residential staff less inclined to individually discuss smoking behaviour. Residential care settings also influence people's ability to maintain healthy routines, as they may both facilitate or hamper healthy lifestyles, such as opportunities for physical activity or healthy diet in group homes [60, 61]. A first step towards recognition of the importance of lifestyle discussions between patients and healthcare professionals is the adequate recording of such conversations.

Strengths and limitations

As we did not have access to full medical records of patients, including medication prescriptions and unstructured textual notes, it is likely we reported an underestimation of frequencies of monitoring of long-term conditions. Tests may have been performed but not recorded in the designated locations in patients' medical records. Nevertheless, registering monitoring of long-term health conditions into standardised medical records safeguards that relevant information can be retrieved easily for healthcare professionals [62]. Additionally, although medication is often an important aspect of disease management, quality of the data only allowed us to examine recording of quality indicators. Van der Heide, Van der Putten [63] reported that 89.0% of people with intellectual disabilities in residential care settings were prescribed medication for long-term health conditions. Our study was therefore only able to reflect part of the reality of disease monitoring. Future research is therefore encouraged to include recordings of medication management.

We also did not have any information on the severity of the intellectual disabilities. This may have impacted our findings, as people with more severe intellectual disabilities often have worse health, worse lifestyle, and increased mortality rates than those with milder intellectual disabilities [64-66]. As guidelines do not consider these differences, and thereby raise questions on the applicability and possibility of lifestyle discussions or clinical examinations, it could have influenced healthcare professionals' decisions for non-compliance to these guidelines.

Another limitation is that underdiagnosis of long-term health conditions in people with intellectual disabilities could have biased our results, meaning that in reality a larger group would have been eligible for inclusion in our sample. Due to the cognitive deficits in people with intellectual disabilities, diagnosing health problems is more difficult, and more often solely based on physical and observable symptoms [50]. For example, distinguishing between asthma and COPD is more complex due to difficulties performing spirometry in people with intellectual disabilities, used to assess COPD [49, 50]. However, we did not have access to medical records of patients with asthma.

Despite these limitations, to the best of our knowledge this study is the first to structurally examine recording of recommended quality indicators for disease monitoring in Dutch residential care settings. By comparing our findings with the baseline of consultations in regular general practice, we highlighted the differences between the general population and the more complex and vulnerable patient population with intellectual disabilities in residential care. By focusing on the population with intellectual disabilities in residential care, we attempted to fill the literature gap regarding management of long-term conditions for this vulnerable patient population. Additionally, we were able to include an 18-month time-period rather than the 12-month time frame recommended by clinical guidelines in which to perform examinations. Sometimes, for practical reasons, intervals of slightly more than twelve months are maintained. Our time frame of 18 months thus reflected reality more closely.

Furthermore, although we only had access to information from one long-term care provider, this organisation operates in 240 municipalities in the Netherlands (about 70% of all municipalities), serving about 13,600 clients [67]. Therefore, it is likely that our findings are generalisable to other care providers. Researchers are invited to shed more light on management of long-term conditions in other residential care settings. In any case, we were able to study a large population of patients with a diagnosis of a long-term health condition, showing an accurate reflection of the recording of quality indicators for disease monitoring in residential care settings.

Implications

The recording of management of long-term conditions for patients with intellectual disabilities in long-term care appears suboptimal. With improved recording of quality indicators for disease monitoring, the course of disease can be better monitored, eventually improving the quality of management of long-term conditions. Non-recording of quality indicators does not mean that long-term health conditions are not monitored. Rather, it may be the case that healthcare professionals experienced more difficulties with either performing clinical examinations in people with intellectual disabilities compared to the general population [47-50], or experienced too high an administrative burden to register indicators performances adequately [52].

Overall, our findings underscore the importance of tailored management for long-term health conditions for patients with intellectual disabilities in residential care. Knowledge and awareness on the differences with the general population, especially on the worse health of this population, allows healthcare professionals to gain more insights into the relevance and necessity of adequately recording quality indicators in medical records. Supplementing guidelines for long-term health conditions with information on these

conditions in people with intellectual disabilities may aid in acquiring this awareness and knowledge. The lack of discussions on healthy lifestyle displays the need for suitable guidelines for patients with intellectual disabilities in residential care settings, and for people with more severe intellectual disabilities. Making residential contexts more healthy by default is promising for the lifestyle of people with ID residing in such contexts.

For both undergraduate students (e.g. medical school) and postgraduate students (e.g. GP trainees) incorporating knowledge on management of long-term health conditions in people with ID within curricula is thus essential for more awareness on patients with intellectual disabilities [68]. Training modules on managing long-term conditions in these patients can aid in them feeling more comfortable in providing care and managing their health conditions.

Although IT systems should support care provision in clinical practice, they do not always seem to meet healthcare professionals' needs [53]. Consequently, important and relevant information might be recorded elsewhere in medical records, resulting in ambiguity regarding how long-term conditions have been managed. It should therefore be explored how administrative burden can be reduced, and how healthcare professionals can be incentivised towards adequate recordings of quality indicators in residential care settings. Eventually, incomplete recordings of clinical examinations might hamper adequate management of long-term conditions, as it limits opportunities for evaluation and improvement. Within a broader trend towards data-driven work methods in healthcare, adequate recording in medical records requires attention from practice, research, and policy.

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APPENDIX

Supplementary Table S1. Description of diagnostic codes for quality indicators, based on 2021 guidelines for cardiovascular disease, diabetes mellitus type 2, and COPD

Description of quality indicator
Quality indicators related to lifestyle
Smoking behaviour ever discussed
Dietary pattern discussed in past 12 months
Physical activity discussed in past 12 months
Alcohol use discussed in past 5 years (2017-2021)
BMI test in past 12 months
Quality indicators related to disease monitoring
Blood pressure test in past 12 months
Glucose test in past 5 years (2017-2021)
eGFR test in past 5 years (2017-2021)
Albuminuria test (albumin/creatinine ratio) in past 12 months
HbA1c test in past 12 months
Foot examination in past 12 months
Fundus exam in past 36 months
Registration of daily functioning in past 12 months (questionnaires CCQ or MRC)
Registration of disease burden in past 12 months
Number of exacerbations registered in past 12 months
Spirometry in past 36 months

Source of codes for quality indicators: based on guidelines of Dutch College of General Practitioners [39-41] and https://bepalingen.nhg.org/labcodes/determinations

	Utilised diagnostic code of indicators
	1814 ADMI, 2039 AFFU, 2033 OAFR, 2011 BASR, 4017 FBHS, 3949 MBSR, 2405 MOSR, 4014 MVBR, 2008 PRSR, 2047 ROAC, 2017 ROBH, 2016 ROIN, 1991 ROJA, 1739 ROOK, 2019 ROTV, 2027 ROVC, 2770 ROVP, 1992 SIPD, 4015 SRBS, 2001 STAF, 2002 STDA, 1996 STOP, 2999 VSNR
	2138 BYVD, 2718 BZVD
	3239 NNGB, 3958 BWRL
	1591 ALCO, 2423 5SHT, 3446 AUDC
	1272 QUET
	3714 RHD7, 3712 RHS7, 3715 RLD7, 3713 RLS7, 2658 RR24, 3337 RR3D, 3336 RR3S, 1852 RRAR, 3327 RRD7, 2669 RRDD, 2056 RRDI, 2188 RRDI, 1740 RRDI, 1741 RRDI, 2668 RRDS, 2660 RRGD, 2659 RRGS, 2662 RRHD, 2661 RRHS, 2664 RRLD, 2663 RRLS, 2667 RRND, 2666 RRNS, 3326 RRS7, 1794 RRSA, 1742 RRSE, 1743 RRSE, 3730 RRST, 3487 RRSW, 2055 RRSY, 2189 RRSY, 1744 RRSY, 1745 RRSY
	2150 GLUC
	1919 KREM, 3740 KREM, 524 KREA, 3583 KREC, 3907 EGFC, 3908 EGCC
	40/42 ALBK
	2816 HBAC
	2196 RIVU, 3609 ZPVU
	2129 FUFO, 1638 DAFU, 3823 DMR7, 3924 DMR7, 1652 DMRP, 1653 DMRP, 3824 FSFU, 3825 DMMA, 3926 DMMA
	2210 MRCD, 2402 CCQT
	3717 ZLC1, 3718 ZLC2, 4035 ZLC2, 3719 ZLC3, 3720 ZLC4, 3013 ZLCP
	3549 COAE
	3647 FVNB, 1677 FVNB, 1678 FVNB

DISEASE MONITORING WITHIN AND OUTSIDE DISEASE MANAGEMENT PROGRAMMES

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ABSTRACT

Introduction: Disease management programmes (DMPs) have been introduced to deliver standardised, high-quality care to patients with chronic diseases. Although chronic diseases are common among people with intellectual disabilities (ID), this approach may be suboptimal for meeting their care needs.

Aim: To examine differences between chronically ill patients with/without ID in DMP enrolment and disease monitoring in Dutch general practice.

Design and Setting: An observational study utilising the Nivel Primary Care Database (2015–2018) comparing patients with ID and cardiovascular disease (CVD), diabetes mellitus (DM), or chronic obstructive pulmonary disease (COPD) with matched (1:5) controls without ID.

Method: Using conditional logistic regression, we examined enrolment in DMP per chronic disease and tested differences between groups in the frequencies of consultations, medication prescriptions, and routine examinations.

Results: We matched 2,653 chronically ill patients with ID with 13,265 controls without ID. DM patients with ID were more likely than controls to be enrolled in DMP (OR=1.44, 95%CI=1.27–1.64). Independent of DMP enrolment, chronically ill patients with ID were more likely than controls to be frequent consulters. DM patients and COPD patients with ID not enrolled in DMPs had more medication prescriptions than non-enrolled patients without ID (OR=1.46, CI=1.10–1.95; OR=1.28; CI=0.99–1.66, respectively). Most patients with ID and their controls enrolled in DMPs received routine examinations at similar frequencies.

Conclusion: Although DMPs do not specifically address the needs of chronically ill patients with ID, these patients do not seem underserved in the management of chronic diseases in terms of consultation, medication, and tests.

INTRODUCTION

To reduce the high impact of chronic diseases, disease management programmes (DMPs) have been introduced in primary care in countries such as Germany, Sweden, United Kingdom, United States, and the Netherlands [1]. DMPs are multidisciplinary efforts to improve the quality and efficiency of chronic disease management by providing continuous, patient-centred, comprehensive care [2, 3]. DMPs encompass the management of patients with high-impact chronic diseases, such as cardiovascular disease (CVD), diabetes mellitus (DM), or chronic obstructive pulmonary disease (COPD) [4]. In the Netherlands, chronic disease management is regionally coordinated by general practice care groups that share responsibility for delivering DMPs to their patients. Quality of care is assured by developing DMPs in line with national disease monitoring standards and by benchmarking practices' performance against national criteria [4], thereby potentially preventing deterioration and complications in chronic diseases while maintaining a high quality of life [5, 6].

For some patient groups, such as patients with intellectual disabilities (ID), reducing the impact of chronic diseases is more complex. Although around 1-1.5% of people globally are diagnosed with ID, the actual prevalence is likely to be higher, as not all IDs are recognised in general practice [7]. People with ID experience significant limitations in intellectual functioning and adaptive behaviour, often manifested as difficulties recognising disease symptoms and understanding disease consequences, and, consequently, have increased health needs and health problems [8-13]. They therefore have to avail of care more than people without ID [8, 14]. Moreover, chronic diseases such as CVD, DM, and COPD are more prevalent among people with ID, develop at younger ages, and occur more often in males with ID than in females with ID [15]. These differences are seldom addressed in existing literature, disease guidelines, or DMPs [16].

Despite these distinct characteristics, chronic disease management for patients with ID is provided mostly within the non-ID-oriented setting of general practice, which can be challenging for general practitioners (GPs). GPs have expressed difficulties in providing care to patients with ID, mostly relating to communication, continuity of care, and time constraints [17]. These difficulties are reflected in compliance with guidelines. Although previous studies do not take DMP enrolment into account, most show that routine examinations advocated in DMPs are provided less frequently for patients with than without ID [18-24]. Non-compliance with protocols and guidelines not only compromises quality of care, but may also contribute to health disparities, and puts chronically ill patients with ID at increased risk of complications, avoidable hospital admissions, and even premature mortality [25, 26].

To tackle these inequalities, it is essential to strengthen the evidence base for providing adequate, suitable care for chronically ill patients with ID. Although DMPs promise quality of care, the complexity of care for patients with ID highlights the need for insights into differences from the general population in DMP enrolment and disease monitoring. This study therefore aimed to examine differences between chronically ill patients with ID and their controls in enrolment in DMPs and disease monitoring in Dutch general practice.

METHOD

Study design and data sources

This observational matched study used the Nivel Primary Care Database (NPCD), which routinely collects information from medical records of over a million patients registered in a representative sample of Dutch general practices [27, 28]. Only patients with a CVD, DM, or COPD diagnosis in their 2018 medical record were selected, using International Classification of Primary Care (ICPC) version 2 (see Supplementary Table S1) [29]. To advance ID identification, we linked population data from Statistics Netherlands to NPCD, as described in more detail elsewhere [7]. This study followed the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines for observational studies [30].

Study groups

For patients with a CVD, DM, or COPD diagnosis, data were collected retrospectively from 2015 onwards, or from diagnosis onwards. Patients with ID were identified by the presence of ICPC code P85 'Mental retardation' in their medical record or when ID indicators were found through data linkage with ID-related social support or long-term care [7]. Selecting all adults (aged 18+) with ID and a chronic disease, we matched them randomly to five people without ID indicators in the same ten-year age group, of the same sex, and with the same chronic disease.

Operationalisations

Patients either with a record of enrolment in a DMP, the GP being coded as main provider for chronic illness, or when >75% of the indicators as advised in the DMP care guidelines were fulfilled [31] were coded as being enrolled in a DMP (Supplementary Figure S1). Patients with solely a CVD diagnosis could be admitted to CVD-DMP, patients with DM (with or without CVD or COPD) to DM-DMP, and patients with COPD (with or without CVD or DM) to COPD-DMP.

To examine disease monitoring, we first examined consultations and medication prescriptions. We calculated the mean of all consultations between 2015 and 2018 for all patients with CVD, DM, and COPD separately (weighted by patients' registration time in general practice). The average (weighted) number of consultations (9.1 consultations per year for CVD, 8.3 for DM, and 9.3 for COPD) was used as cut-off point to distinguish between less and more frequent consulters. Similar calculations were used for medications based on prescriptions for CVD, DM, or COPD. The average (weighted) number of prescriptions between 2015 and 2018 for all patients with CVD, DM, or COPD (1.8 prescriptions per year) was used as cut-off point to distinguish between less and more frequent users.

Second, we examined frequency of key routine examinations in 2018. Several indicators considered essential for the monitoring of chronic disease according to Dutch and international guidelines were found [4, 21, 32, 33]. For CVD, these were presence of a record of LDL measurement for those below 80 years of age and of blood pressure test. For DM, these were presence of a record of HbA1c measurement and of albuminuria test. For COPD, these were record of smoking behaviour and having at least two prescriptions for inhalation medication (R03A or R03B).

Statistical analysis

Descriptive statistics of the study groups were presented as frequencies with percentages or in means with standard deviations. Frequencies with percentages were presented for the proportion of patients enrolled in DMPs, consultations, medication prescriptions, and adherence to routine examinations. Information on the latter three was stratified by enrolment in DMPs. Differences between patients with ID and controls were compared using conditional logistic regression, estimating odds ratios (ORs) and 95% confidence intervals (CIs). *P*-values below 0.05 were considered statistically significant. All analyses were conducted in SPSS (version 25.0).

RESULTS

Demographics

Patients with ID and at least one chronic illness (n=2,653) were identified and matched with 13,265 controls without ID (Table 1). Most patients with and without ID were male (57.3%) and were between 51 and 70 years old (58.5%). Most patients had a diagnosis of DM (59.7%), followed by CVD (31.9%) and COPD (27.7%). Of patients with ID, 62.0% were registered at least three years at the GP compared to 59.3% of controls. Of patients with ID, 70.2% (n=593) had an indication for CVD-DMP, compared to 69.8% (n=2,952) of controls.

Table 1. Descriptive statistics of patients with and without intellectual disabilities (ID) with chronic diseases N (%)

	Patients with ID	Controls
Total	2,653	13,265
Sex		
Males	1,519 (57.3)	7,595 (57.3)
Females	1,134 (42.7)	5,670 (42.7)
Mean age (SD)	54.7 (13.7)	55.2 (13.9)
Age		
18–30 years	193 (7.3)	965 (7.3)
31–50 years	644 (24.3)	3,220 (24.3)
51–70 years	1,553 (58.5)	7,765 (58.5)
71 years or older	263 (9.9)	1,315 (9.9)
Registration time at GP		
≤1 year	370 (13.9)	1,751 (13.2)
1.1–2 years	468 (17.6)	2,834 (21.4)
2.1–3 years	169 (6.4)	819 (6.2)
3.1–4 years	1,646 (62.0)	7,861 (59.3)
Diagnosis of chronic disease[1]		
CVD	845 (31.9)	4,228 (31.9)
Indication for CVD-DMP[2]	*593 (70.2)*	*2952 (69.8)*
DM	1,584 (59.7)	7,575 (57.1)
Indication for DM-DMP[2]	*1,584 (100.0)*	*7,575 (100.0)*
COPD	736 (27.7)	3,680 (27.7)
Indication for COPD-DMP[2]	*736 (100.0)*	*736 (100.0)*

[1] Prevalence of chronic diseases is estimated with diagnoses in medical records based on International Classification of Primary Care (ICPC) codes.

[2] If patients had diabetes mellitus as well as cardiovascular disease, they were admitted to DM-DMP. Those with only cardiovascular disease could be admitted to CVD-DMP. Of those with ID, 593 had an indication for CVD-DMP, compared to 2,952 of controls without ID.

Enrolment in DMPs

Enrolment of people with ID in DMPs for CVD (43.8%) and COPD (41.0%) was comparable and not statistically significantly different from controls (Figure 1). DM patients with ID were more likely to be enrolled in a DMP than controls (69.8% vs 62.6%; OR=1.44, 95%CI=1.27–1.64; Supplementary Table S2), and received DM care in specialist care settings less often than their controls (4.9% vs. 7.0%; OR=0.65, CI=0.51–0.84).

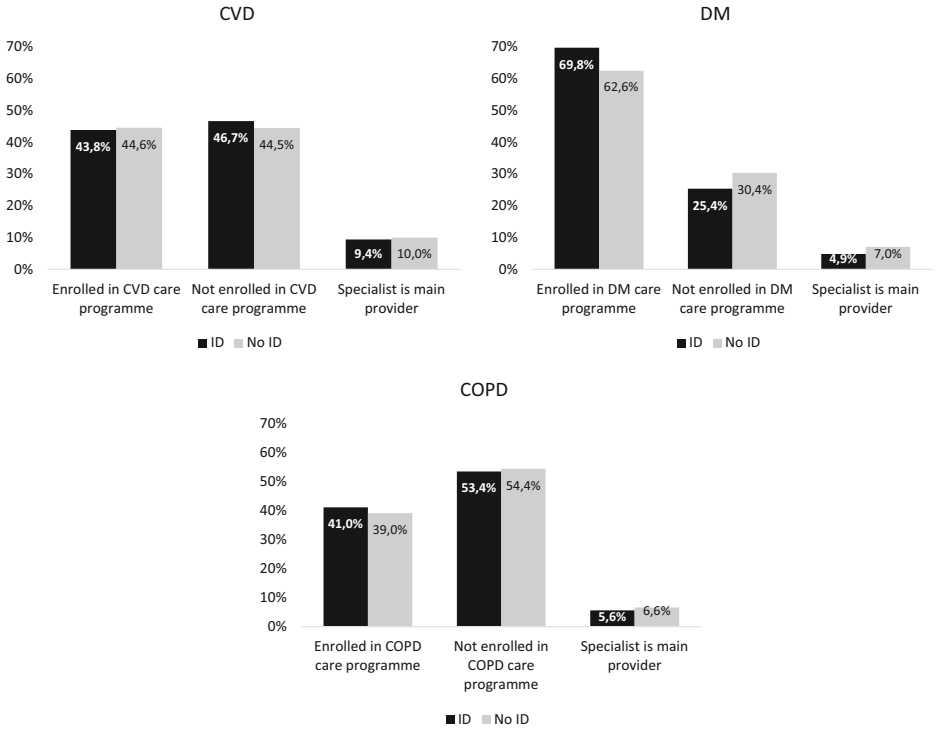

Figure 1. Distribution of patients with and without intellectual disabilities (ID) by type of management for their chronic illness

Disease monitoring

For those enrolled in DMPs, chronically ill patients with ID were more likely than their controls to be frequent consulters: that is, for patients with ID and CVD (OR=2.71, CI=1.91–3.84), DM (OR=2.49, CI=2.14–2.89), and COPD (OR=3.01, CI=2.18–4.16; Table 2). Similar results were found for those not enrolled in DMPs (Table 2).

On average, between 2015 and 2018, patients with CVD, DM, and COPD received 1.8 prescriptions per year. Only among those not enrolled in DMPs did we observe differences in frequency of medication prescriptions: patients with ID and DM (OR=1.46, CI=1.10–1.95) or COPD (OR=1.28, CI=0.99–1.66) were more likely to receive ≥1.8 prescriptions per year than their controls (Table 2).

In those enrolled in CVD-DMP, there were no significant differences between patients with ID and their controls in the frequency of LDL checks (resp. 92.0% vs. 92.3%) and blood pressure tests (78.8% vs. 78.0%). In those not enrolled in CVD-DMP, patients with ID had a lower likelihood (30.1%) than their controls (41.1%; OR=0.60, CI=0.42–0.87) of receiving an LDL check (Table 2).

Table 2. Frequency and percentage of chronically ill patients with and without intellectual disabilities (ID) on disease monitoring within and outside disease monitoring programmes (DMPs) in 2015-2018[a]

Cardiovascular disease	
Frequent consulters (≥9.1) per year, N (%)[c]	
Frequent CVD medication prescriptions (≥1.8) per year, N (%)[d]	
LDL measurement (only those <80 years) in 2018	
Blood pressure test in 2018	
Diabetes mellitus	
Frequent consulters (≥8.3) per year, N (%)[c]	
Frequent DM medication prescriptions (≥1.8) per year, N (%)[d]	
HbA1c measurement in 2018	
Albuminuria test in 2018	
COPD	
Frequent consulters (≥9.3) per year, N (%)[c]	
Frequent COPD medication prescriptions (≥1.8) per year, N (%)[d]	
At least 2 prescriptions of inhalation medication (R03A/B) in 2018 (only those with inhalation medication prescriptions)	
Smoking behaviour discussed in 2018	

** p<0.005, * p<0.05

[a] The 2018 DMP guidelines state that all monitoring tests should take place once a year.

[b] OR=conditional odds ratio; CI=confidence interval

[c] Weighted average (by patients' registration time in general practice) of number of contacts between

There were no significant differences between DM patients with ID and their controls enrolled in the DMP in the frequency of HbA1c (resp. 90.5% vs. 88.7%) and albuminuria measurements (resp. 77.5% vs. 79.1%). HbA1c was more likely to be measured in DM patients with ID not enrolled in the DMP (36.8%) than their controls (24.8%; OR=1.85, CI=1.34-2.55; Table 2).

In COPD patients with and without ID enrolled in COPD-DMP, occurrence of receiving 2+ prescriptions of inhalation medication (resp. 79.1% and 79.2%) and discussing smoking behaviour with care providers (resp. 80.5% and 78.1%) was similar. COPD patients with ID not enrolled in COPD-DMP were more likely to receive two or more prescriptions of inhalation medication (80.8%) than their controls (69.5%; OR=1.84, CI=1.32-2.57; Table 2).

Enrolled in DMP			Not enrolled in DMP		
Patients with ID	Controls	OR (95% CIs)[b]	Patients with ID	Controls	OR (95% CIs)[b]
N=260	N=1,316		N=277	N=1,340	
130 (50.0)	371 (28.2)	2.71** (1.91; 3.84)	111 (40.1)	360 (26.9)	1.78** (1.28; 2.49)
69 (26.5)	354 (26.9)	0.91 (0.62; 1.32)	88 (31.8)	388 (29.0)	1.02 (0.73; 1.43)
230/250 (92.0)	1,170/1,268 (92.3)	0.80 (0.43; 1.49)	81/269 (30.1)	531/1,293 (41.1)	0.60* (0.42; 0.87)
205 (78.8)	1,027 (78.0)	0.96 (0.65; 1.42)	81 (29.2)	406 (30.3)	1.01 (0.72; 1.41)
N=1,105	N=4,739		N=402	N=2,302	
607 (54.9)	1,582 (33.4)	2.49** (2.14; 2.89)	194 (48.3)	620 (26.9)	2.83** (2.05; 3.89)
348 (31.5)	1,403 (29.6)	1.07 (0.91; 1.25)	159 (39.6)	751 (32.6)	1.46* (1.10; 1.95)
1,000 (90.5)	4,205 (88.7)	1.20 (0.94; 1.53)	148 (36.8)	571 (24.8)	1.85** (1.34; 2.55)
856 (77.5)	3,747 (79.1)	0.88 (0.74; 1.05)	115 (28.6)	464 (20.2)	1.43 (0.99; 2.07)
N=302	N=1,437		N=393	N=2,001	
174 (57.6)	490 (34.1)	3.01** (2.18; 4.16)	198 (50.4)	554 (27.7)	2.88** (2.22; 3.73)
99 (32.8)	424 (29.5)	1.07 (0.78; 1.46)	137 (34.9)	586 (29.3)	1.28* (0.99; 1.66)
110/139 (79.1)	519/655 (79.2)	1.04 (0.78; 1.39)	80/99 (80.8)	282/406 (69.5)	1.84** (1.32; 2.57)
243 (80.5)	1,123 (78.1)	1.32 (0.89; 1.95)	61 (15.5)	268 (13.4)	1.36 (0.96; 1.94)

patients and general practitioners (e.g., regular consultations, home visits) between 2015 and 2018. For CVD, this average was 9.1 consultations per year, for DM 8.3 per year, and for COPD 9.3 per year.
[d] Weighted average (by patients' registration time in general practice) of medication prescriptions between 2015 and 2018 across the study populations were 1.8 prescriptions per year for CVD, DM, and COPD.

DISCUSSION

Summary

This study compared enrolment in DMPs and disease monitoring for chronically ill patients with ID and their controls. Only DM patients with ID were more often enrolled in DMPs. Patients with ID enrolled in CVD-DMP, DM-DMP, or COPD-DMP received consultations more often than their enrolled controls. Among those not enrolled in a DMP, patients with ID received consultations, medication prescriptions, and routine examinations (apart from LDL measurement) more often than their controls. Despite the frequently reported care inequities between people with and without ID, we did not find evidence for limited access to DMPs for patients with an ID diagnosis and a chronic disease. Additional research is needed to explore whether chronic disease management meets the needs of this complex patient population.

Comparison with existing literature

To the best of our knowledge, DMP enrolment for patients with and without ID has not yet been studied, making comparison difficult. Our finding that patients with ID are often enrolled in DM-DMP could reflect their increased disease burden, as DMP enrolment is often higher among those with a higher disease burden and of younger ages [33-35]. However, future research could further examine the influence of socioeconomic determinants of enrolment in people with and without ID.

No studies have been identified that examined disease monitoring by DMP enrolment for patients with ID. Disregarding DMP enrolment, similar to previous studies, we found that patients with ID have higher consultation rates than those without ID, and in general have high medication use [8, 36-38]. Although medication in people with ID may be prescribed more often to regulate challenging behaviour [39, 40], we were unable to consider medication type in our analyses.

Our finding that the frequency of routine examinations did not differ between those with and those without ID conflicts with previous research. Most studies showed fewer tests in people with ID compared to the general population. Often, ID patients with CVD were found to receive fewer cholesterol and blood pressure tests, those with DM received microalbuminuria or HbA1c tests less often, and those with COPD discussed their smoking status or had smoking cessation advice less often than their controls [19, 20, 22, 24]. Several reasons can be proposed for these seemingly contradictory findings. First, perhaps our novel focus of investigating DMP enrolment before examining monitoring accounted for these differences: it could be that the financial incentives to stimulate DMP enrolment ensure increased compliance with chronic disease guidelines. Second, by innovatively linking administrative data to medical records, we provided a more thorough identification of the people with ID group. However, future research should further investigate these differences.

Among those not enrolled in DMPs, we found that most tests for routine examinations were more often administered in ID patients. Nevertheless, patients with (and without) ID not enrolled in DMPs still received fewer routine examinations than those enrolled. To the best of our knowledge, no previous studies examined similar research questions. Research on patients' experiences and outcomes is therefore required to interpret findings on routine examinations within and outside DMPs for patients with ID.

Strengths and limitations

This study is the first to examine DMP enrolment and disease monitoring for patients with ID and their controls by DMP enrolment using large-scale, individual-level data from a population-based primary care database. By linking datasets and using multiple methods to identify people with ID, we reduced the common problem of underreporting ID in medical records [41] and could make more robust claims about a larger group of patients with ID than is possible by using solely GP records [7, 41].

Thanks to our matched study groups, we limited influences of sociodemographic factors and acknowledged the different demography of the ID population [7]. As previous research has shown that these factors are important in the prevalence of chronic diseases and care utilisation in people with ID, as well as enrolment in DMPs [15, 34, 36, 42-44], we explored DMP enrolment and disease monitoring without their interference.

Additionally, by utilising multiple methods to asses (non-)enrolment in DMPs, we reduced the risk of wrongly assigning patients and provided a more precise image than by focusing solely on one method. The NPCD, containing information directly derived from clinical practice, allowed us to examine enrolment and disease monitoring as objectively as possible. However, our findings do not fully comply with the 2018 benchmark from the national primary care organisation InEen. Unlike NPCD, this benchmark contains solely information from general practices, which are part of regional care groups [45]. Future research should therefore consider regional differences.

Besides insufficient registration for DMP enrolment, registration for routine examinations and medication prescriptions is also most likely incomplete. Perhaps care providers registered routine examinations and medication prescriptions more thoroughly for those enrolled in DMPs because of DMP-related financial incentives [4]. Although 85% of the total study population had no medication prescriptions, missing data occurred at a similar frequency for people with and without ID.

Implications

This study is the first to explore DMP enrolment and disease monitoring in the Netherlands for chronically ill patients with and without ID. Although previous studies on disease monitoring among patients with ID often reveal a rather unfavourable image for these patients compared to the general population, this study shows that focusing on DMP enrolment can reveal other patterns: both patients with and patients without ID benefit hugely from DMP enrolment. As DMPs often improve clinical outcomes and quality of care for chronically ill patients [33, 46, 47], policymakers should encourage healthcare insurers and care providers to provide suitable (financial) incentives for

enrolling patients in DMPs [35]. Patients with ID not enrolled in DMPs should be monitored continuously, as their already poorer health might further deteriorate without adequate recognition of health conditions [48]. With their strong generalist skills, GPs have a solid foundation to address ID patients' complex healthcare needs [48]. Despite the similarities in enrolment, awareness of patients' individual care needs is thus an essential starting point for equitable and suitable chronic disease management [15, 49].

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APPENDIX

Supplementary Table S1. Selection of four chronic diseases and accompanying ICPC codes

Chronic disease	ICPC code
Ischaemic heart disease	
Angina pectoris	K74
Myocardial infarction	K75
Other/chronic ischemic heart disease	K76
Cerebrovascular disease	
Transient ischemic attack	K89
Cerebrovascular accident	K90
Diabetes mellitus	T90
Chronic obstructive pulmonary disease	
Chronic bronchitis/bronchiectasis	R91
Emphysema/COPD	R95

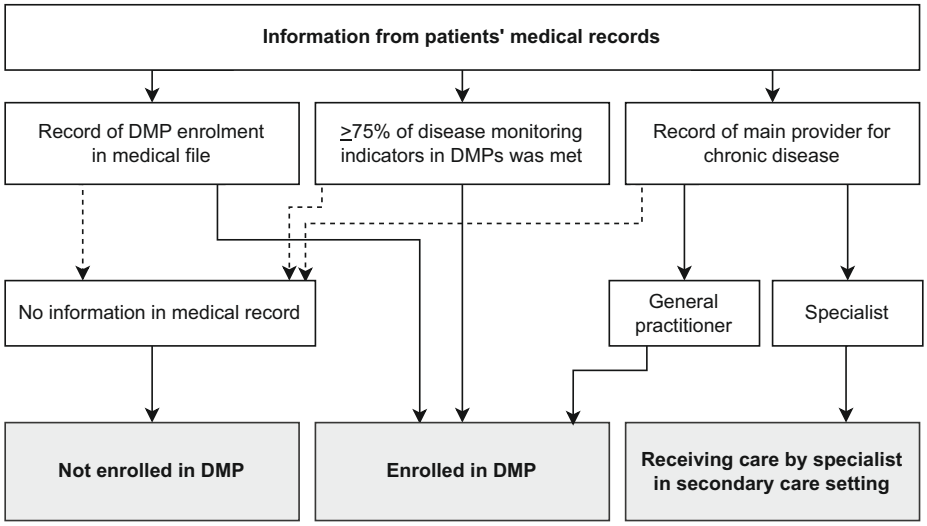
Supplementary Table S2. Distribution of chronically ill patients with and without intellectual disabilities (ID) (not) enrolled in disease management programmes (DMPs)

	Cardiovascular disease[a]	Diabetes mellitus[a]	COPD
Enrolled in DMP			
Patients with ID	260/593 (43.8)	1,105/1,584 (69.8)	302/736 (41.0)
Controls	1,316/2,952 (44.6)	4,739/7,575 (62.6)	1,437/3,680 (39.0)
ORs[b] (95% Cis)	0.96 (0.79; 1.17)	**1.44** (1.27; 1.64)**	1.09 (0.93; 1.29)
Not enrolled in DMP			
Patients with ID	277/593 (46.7)	402/1,584 (25.4)	393/736 (54.4)
Controls	1,340/2,952 (45.4)	2,302/7,575 (30.4)	2,001/3,680 (54.4)
ORs[b] (95% Cis)	1.06 (0.87; 1.29)	**0.77** (0.68; 0.88)**	0.96 (0.81; 1.13)
Care provision in secondary care setting			
Patients with ID	56/593(9.4)	77/1,584 (4.9)	41/736 (5.6)
Controls	296/2,952(10.0)	534/7,575(7.0)	242/3,680(6.6)
ORs[b] (95% Cis)	0.95 (0.69; 1.31)	**0.65**(0.50; 0.84)**	0.84 (0.59; 1.18)

* p<0.05, ** p<0.005 (significant results also shown in bold)

[a] If patients had diabetes mellitus as well as cardiovascular disease, they were admitted to DM-DMP. Those with only cardiovascular disease could be admitted to CVD-DMP. Of those with ID, 593 had an indication for CVD-DMP, compared to 2,952 of controls without ID.

[b] OR=conditional odds ratio; CI=confidence interval



Supplementary Figure S1. Indicators used to assess enrolment in disease management programme (DMP) or receiving care in secondary care setting

PART III

CARE NEEDS

CARE NEEDS OF CHRONICALLY ILL PATIENTS: PATIENTS' AND PROVIDERS' PERSPECTIVES

ABSTRACT

Introduction: To reduce the impact of chronic diseases (cardiovascular disease, diabetes mellitus type 2, and chronic lung disease (asthma or chronic obstructive pulmonary disease (COPD)), it is imperative that care is of high quality and suitable to patients' needs. Patients with intellectual disabilities (ID) differ from the average patient population in general practice because of their limitations in adaptive behaviour and intellectual functioning, and concomitant difficulties recognising and reacting to disease symptoms, proactively searching health information, and independently managing diseases effectively. Because of these differences, information on their care needs is essential for suitable chronic disease management (CDM). Inadequate recognition of the care needs of this vulnerable population may hamper the harmonisation of evidence-based and person-centred care, compounded by issues such as stigma, misconceptions, and diagnostic overshadowing. This study therefore aimed to explore the needs of patients with ID and of healthcare providers (HCPs) in the context of CDM in general practice.

Methods: This qualitative study recruited patients with ID for face-to-face individual interviews and HCPs for focus groups. With the Chronic Care Model as the underlying framework, semi-structured interviews and focus-group guides were defined to explore patients' care needs and HCPs' perspectives. All interviews and focus groups were audio-recorded and transcribed verbatim. Using Atlas.ti software, data were analysed using thematic analysis.

Results: Between June and September 2022, 14 patients with ID and cardiovascular disease, diabetes mellitus type 2, and/or asthma/COPD were interviewed; and 32 general practitioners and practice nurses participated in seven focus groups. We identified six care needs underpinning suitable CDM: trusting relationship between patient and HCP; clear expectations about the CDM process; support in disease management; directive decision-making; support in healthy lifestyle; accessible medical information.

Conclusions: This vulnerable patient population has complex care needs that must be acknowledged for suitable CDM. Although HCPs largely recognised these needs, organisational factors and lack of training or experience with patients with ID hampered HCPs' ability to fully adjust care provision to these needs. Access to, and knowledge of, easy-language information on chronic diseases and communication guidelines could aid HCPs to facilitate patients in managing their diseases more adequately.

INTRODUCTION

To reduce the high impact of chronic diseases such as cardiovascular disease (CVD), diabetes mellitus (DM) type 2, and chronic lung disease (asthma/and or chronic obstructive pulmonary disease (COPD), it is imperative that care is of high quality and suitable to patients' care needs [1]. To ensure 'the most appropriate care at the most appropriate time and place in the most efficient manner' [2], chronic disease guidelines support healthcare providers (HCPs) in delivering this care. However, principles of evidence-based medicine and person-centred medicine may yield different views on what constitutes most appropriate care (one-size-fits-all versus personalised approach, respectively) [3, 4], in particular when it concerns patient groups that differ in care needs from the average patient population.

One patient group that may require a different approach in healthcare is that of patients with intellectual disabilities (ID). Although the prevalence of ID is estimated at 1.5% in Western countries, people with ID are overrepresented in chronic disease groups, such as CVD, DM, and asthma/COPD [5, 6]. Limitations in adaptive behaviour and intellectual functioning mean that people with ID often have difficulty recognising and reacting to disease symptoms, proactively searching health information, retaining information from their HCP, and independently managing diseases effectively [7-9]. As a result, such patients often require easy-language information, support in utilising healthcare, and increased health surveillance [10-14].

These care needs have been identified mainly in contexts with aims other than chronic disease management (CDM) (e.g., palliative, hospital, outpatient, or social care [10-14]). Because of the perpetuity of CDM, and to prevent comorbidities and exacerbations, understanding care needs in the CDM context is essential. Inadequate recognition of the care needs of this vulnerable population may hamper the harmonisation of evidence-based and person-centred care, compounded by issues such as stigma, misconceptions, and diagnostic overshadowing, where symptoms are wrongly attributed to the ID rather than to health problems [15, 16]. Consequently, health(care) inequities between people with and without ID continue to exist. This study therefore aimed to explore the needs of patients with ID and of HCPs in the CDM context for patients with ID.

METHODS

Design and context

This study is qualitative, combining views of patients with ID and HCPs (general practitioners (GPs) and practice nurses (PNs)). Semi-structured individual interviews yielded an in-depth understanding of the personal experiences of chronically ill patients with ID. The focus-group setting allowed for broad exploration of HCPs' perspectives by sharing experiences in providing care to chronically ill people with ID. Interviews and focus groups were conducted non-sequentially. The study protocol was preregistered (https://osf.io/b4er7).

In the Netherlands, general practice (GPs and PNs) plays a pivotal role in managing chronic diseases for the majority of patients with (and without) ID, offering accessible and comprehensive care [5, 17, 18]. PNs take up most tasks of chronic disease management, such as monitoring disease progression, patient education, and signalling complications. Their active involvement in chronic disease management has been shown to increase quality of care [19-21]. The Chronic Care Model, the foundation for national care standards that specifies prerequisites for high-quality CDM [22, 23], served as the basis for the interview and focus-group guides (Figure 1).

Study populations and recruitment

Two distinct populations of study participants were recruited between January and September 2022. Persons with ID were recruited who were 18 years or older, had a chronic disease (i.e., CVD, DM, and/or asthma/COPD) for which they actively received CDM, and could communicate verbally in an interview. People with borderline, mild, or moderate ID were recruited, because these groups are most likely to be able to be valuable interviewees [24]. Patients were recruited via GPs, advocacy groups for people with ID, care organisations, and snowball methods. GPs and PNs were recruited who provided CDM to patients with (suspected) ID. They were approached via flyers, email, face-to-face, education days for GPs, and snowball methods [25].

Purposive sampling was used to reflect variation in perspectives and backgrounds [26] in order to construct a holistic understanding of CDM [27]. Practically, this meant that we selected male and female patients of various ages with various chronic diseases (i.e., CVD, DM, asthma/COPD) living in various settings (i.e., in residential-care organisations, individually, or with family). In addition, we selected male and female HCPs of various ages with various experience in providing CDM to patients with ID.

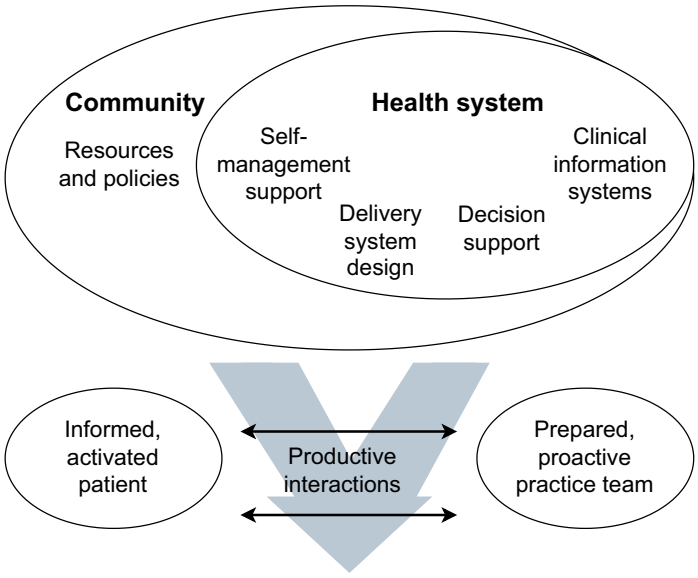


Figure 1. Chronic Care Model

Figure reproduced from Wagner [22]. The Chronic Care Model posits that six domains are the foundation of chronic care. In the *health system*, the structure, goals, and values should revolve around providing high-quality care to patients. Second, *self-management support* in the sense of patient education helps patients and relatives to acquire skills to manage the chronic disease adequately. Third, to aid healthcare providers (HCPs) with *decision support*, it is essential that evidence-based clinical guidelines are incorporated into practice. Fourth, the way in which *delivery systems* are designed, for instance in multidisciplinary teams, can make care more efficient. Fifth, adequate *clinical information systems* may improve compliance to guidelines or care planning. Sixth, *community resources*, such as linkages with other HCPs or community-based resources may aid in short lines of communication, through which carers and HCPs may cooperate efficiently.

All study participants had to read and sign an informed consent form before participating in their interview or focus group. This study followed international guidelines for reporting qualitative research (COREQ) [28].

Data collection

Semi-structured interview and focus-group guides (see appendix) were developed based on the Chronic Care Model and literature on care experiences of people with ID. The interview guide was developed in collaboration with a co-researcher with ID and pilot-tested in two chronically ill patients with ID. The focus-group guide was tested in a group of HCPs to ensure that no relevant themes were missed.

The first author (MvdB) conducted the interviews and moderated the focus groups, after receiving relevant training. Interviews were held face-to-face, and, if required, in the presence of a third person to help the participant answer questions or add relevant information. Focus groups were held during pre-planned education days at the university medical centre (Radboudumc) or online. The first two focus groups were observed by an assistant, who took notes and observed the atmosphere in the groups.

All interviews and focus groups (see Table 1) were audio-recorded and transcribed verbatim. When inductive thematic saturation was achieved during analysis – meaning that no new codes or themes emerged – data collection was completed.

Data analysis

The data were analysed by thematic analyses using Atlas.ti software, whereby we identified common themes in the data through consecutive coding steps [29]. The first five interviews were analysed independently by MvdB, MK, and BS to iteratively shape a preliminary code tree using the predefined themes from the Chronic Care Model underlying the interview guide, while allowing additional themes to emerge. After five interviews, consensus was reached, and the remaining nine interviews were coded independently by MvdB and either MK or BS, further refining the code tree. Preliminary themes were then formulated. Consecutively, the focus groups were similarly coded and analysed. The first focus group was coded by MvdB, MK, and BS. The remaining focus groups were coded independently by MvdB and either MK or BS, and discussed afterwards, leading to preliminary themes resulting from the focus groups.

The preliminary themes from both the interviews and the focus groups were combined and reshaped into overarching themes. The final six overarching themes were defined as care needs, in which we were able to include all relevant information emerging from the data. After discussion and agreement by all authors, the themes were written down and supported by relevant quotes.

RESULTS

Between June and September 2022, 14 individual face-to-face interviews were held with patients with ID and CVD (n=7), asthma/COPD (n=6), and/or DM (n=4). We interviewed eight males and six females, aged 19–74 years old (mean age 47.3 years; Table 1). In three interviews, a third person was present. Additionally, one focus group with PNs (N=6) and six focus groups with GPs (N=26; 3–7 participants per group) were conducted. The groups included 14 males and 18 females (Table 1), aged 29–68 years (mean age 47.3 years).

We identified six overarching care needs for patients (Table 2): 1) trusting relationship; 2) clear expectations about the CDM process; 3) support networks in CDM; 4) directive decision-making; 5) support in healthy lifestyle; 6) accessible medical information. We discuss these needs from the perspectives of patients, GPs, and PNs.

1. Trusting relationship between HCP and patient

Both patients and HCPs stated that a trusting mutual relationship was essential for patients' CDM. Without a trusting relationship, the other identified care needs could often not be achieved.

Patients

For most patients, a trusting relationship was primarily determined by HCPs' use of language:

> *We have a good relationship with our GP and with the cardiologist, it's fine simply because they speak in easily understandable language.* (P11)

The feeling of being taken seriously benefits long-term patient–HCP relationships. Patients indicated that this was mostly determined by HCPs' listening skills, availability (in terms of time and responsiveness), and ability to reassure patients. Some patients explicitly mentioned their ID to their HCP to avoid difficult language; others suggested that they felt taken more seriously when HCPs knew about their ID diagnosis. Patients often felt safer during consultations when HCPs addressed their daily life (such as their hobby) before medical aspects. This trusting relationship facilitated information transfer for patients.

HCPs

Most HCPs recognised the importance of continuity of care, as it functioned as a precondition for a trusting relationship to develop. For most, a trusting relationship started with recognising the ID. In almost all focus groups, without prompts, participants first shared their difficulties experienced in timely recognition of ID, before reflecting on CDM for these patients.

Table 1. Description of interview and focus-group participants

Interviews			
No	*Duration*	*Sex*	*Age*
1	37m	Female	29
2	66m	Female	56
3*	48m	Female	53
4	45m	Male	69
5	39m	Female	42
6	29m	Male	59
7	41m	Male	52
8	34m	Female	19
9	36m	Male	32
10	28m	Male	33
11*	33m	Male	52
12*	38m	Female	40
13	23m	Male	74
14	31m	Male	52
Focus groups			
No	*Duration*	*Sex*	*Age range*
FG1 (GPs)	68 min	4 (2 M, 2 F)	29-46
FG2 (GPs)	62 min	5 (1 M, 4 F)	46-66
FG3 (GPs)	67 min	7 (3 M, 4 F)	38-55
FG4 (GPs)	53 min	4 (4 M, 1 F)	37-49
FG5 (GPs)	71 min	3 (3 M)	32-68
FG6 (GPs)	57 min	3 (2 M, 1 F)	39-63
FG7 (PNs)	59 min	6 (6 F)	38-60

GP = general practitioner; PN = practice nurse; ID = intellectual disabilities; * = third person present at interview.

Living situation	*Chronic disease*	*Self-reported experience with ID*
Independent living, ambulatory care	Chronic lung disease	All recently graduated; little experience with patients with ID
Independent living, ambulatory care	Cardiovascular disease	Most (3/5) provide care to few patients with (suspected) ID
Group home	Cardiovascular disease, diabetes mellitus	Most (5/7) provide care to many patients with (suspected) ID
Independent living, no care	Cardiovascular disease, chronic lung disease	All (4/4) provide care to some patients with (suspected) ID
Independent living, no care	Chronic lung disease	All (3/3) provide care to some/many patients with (suspected) ID
Group home	Cardiovascular disease	All (3/3) provide care to some/many patients with (suspected) ID
Group home	Diabetes mellitus	All (6/6) provide care to many patients with (suspected) ID
Group home	Cardiovascular disease, chronic lung disease	
Independent living, ambulatory care	Cardiovascular disease	
Independent living, ambulatory care	Chronic lung disease	
Independent living, ambulatory care	Cardiovascular disease	
Independent living, ambulatory care	Diabetes mellitus	
Group home	Diabetes mellitus	
Group home	Chronic lung disease	

Table 2. Summary of care needs according to patients and healthcare providers (HCPs)

Theme	Explanation of theme
Trusting relationship between HCP and patient	A trusting relationship was an essential precondition for patients' chronic disease management.
Clear expectations about the disease management process	Patients required clarity before, during, and after disease monitoring consultations.
Support networks in assisting with disease management	Patients' formal and informal support networks played important roles in managing patients' chronic diseases.
Directive decision-making processes	Patients expected HCPs to make decisions for them, but this approach was contradictory to HCPs' preferences.
Support network to assist in achieving and maintaining a healthy lifestyle	Patients and HCPs acknowledged patients' dilemma around independence versus support needed to achieve and maintain healthy habits.
Accessible medical information	For patients and HCPs, medical information should be accessible to benefit continuity of care.

ID = intellectual disabilities; HCP = healthcare provider; CDM = chronic disease management.

	Patients	HCPs
	A trusting relationship was determined by HCPs' use of language: using simple words, and addressing not just patients' medical needs but them as a person. To enforce simple language, some patients explicitly disclosed their ID.	A trusting relationship started with HCPs recognising the ID, as it indicated that they should adapt their language and approach. They felt like they should address patients' daily life before addressing medical needs. Patients' previous negative (care) experiences could make it more complex to build a relationship.
	Unclear arrangements or unmet expectations caused feelings of stress and frustration.	HCPs did not always explicitly discuss expectations or assumed arrangements were clear. Although they attempted to meet expectations, to do so they required knowledge on adequate approaching and the needs of patients.
	Support networks were necessary control mechanisms and provided patients with reassurance. Carers often coordinated access to care and assisted with information transfer between patients and HCPs.	HCPs often relied on carers for relevant information on patients' chronic disease, and for (re-)explaining information to patients in more suitable language. It was difficult to gain an overview of patients' support networks and each one's roles and responsibilities in CDM.
	The higher the HCP in the subjective medical hierarchy, the more patients valued and followed decisions and advices. Although included in decision-making processes, patients expected HCPs to make final decisions.	PNs experienced difficulties because patients valued their advices less than those of GPs. Most HCPs applied a more paternalistic approach and set smaller goals to get patients to value and follow advices.
	Patients aspired to independence but acknowledged they required support to make healthy choices. Small lifestyle modifications were seen as great accomplishments. Their living environment could either hamper or stimulate healthy lifestyles.	Bigger lifestyle modifications were necessary for patients according to HCPs, as part of adequate CDM. They attempted to mobilise support networks as much as possible to achieve healthy lifestyles.
	It could be frustrating and confronting for patients to have to repeat their medical history to different HCPs. Online medical files were useful for checking with carers.	Collaboration between care organisations was difficult with inaccessible medical information in different medical records. Determining the contents of treatment plans for chronic diseases was more complex.

A diagnosis of ID in patients' medical records functioned as an important signal to adapt language accordingly. HCPs mentioned that they adjusted their communication by speaking calmly, using informal language, and keeping sentences short. They also used other adapted approaches: scheduling longer consultations, incorporating humour, and discussing patients' daily life before addressing medical needs. HCPs considered patients' negative past care experiences challenging, although these approaches helped them foster connections and create an environment for trust to develop. This allowed patients to feel at ease and share medical information more easily:

> *With them [patients you've known longer] you know a little bit about their lives so you can comment on them, like 'how's your hobby?'. They all often have fun things that they do in their free time. You know that, so it's easier to make connections or bonding. … Once you have the trust [of patients], those contacts are often much easier in terms of communication … in terms of trust that you're there to help them.* (GP1, FG4)

2. Clear expectations about the CDM process

Most patients required clarity and predictability before, during, and after disease monitoring consultations (e.g., starting at the agreed time or having to take medication at home), although HCPs did not always explicitly discuss what patients could expect.

Patients

Patients often talked about their need for clarity. Unclear arrangements or unmet expectations could result in feelings of stress and frustration:

> *[Name] had told me that the agreement was that I'd get the results of the bloodwork on Friday. … Then I was suddenly called on Wednesday. And then you start to worry. … Sorry, but then I get snappy. … Because then I don't feel so much anger but frustration, … that you were worried for two days extra although nothing was wrong.* (P5)

Patients often expected clinical examinations (e.g., blood pressure measurements) to be performed during CDM check-ups. These examinations reassured them about their self-management skills. When a consultation unexpectedly did not include such tests, patients often failed to see the relevance of that appointment, causing feelings of frustration.

HCPs

Unlike patients, HCPs deemed consultations focused solely on conversation, without clinical examinations, adequate to gain information about patients' CDM. However, HCPs monitored patients with ID more closely than other patients, by scheduling more frequent consultations and conducting clinical examinations or health checks more frequently.

> *When taking someone else's blood pressure, you say 'send the results'. With this one you say: 'Come back in a week to measure your blood pressure again, make an appointment.'* (GP4, FG2)

To meet patients' expectations as well as possible (e.g., performing clinical examinations during consultations, simple-language explanation of CDM), HCPs acknowledged the need for knowledge and experience in working with patients with ID. They therefore expressed the wish for accessible training and sharing thoughts with more experienced colleagues.

3. Support networks in assisting with CDM

(In)formal support networks (e.g., carers, family, care providers) had important roles in CDM at home and during consultations, even though both patients and HCPs found it difficult to identify actors involved and the responsibilities that they had in these networks.

Patients

Carers served as the primary source for patients' questions about their chronic disease, jointly deciding whether to consult the internet or the GP. Most patients indicated that their support networks functioned as a control mechanism and reassurance to check whether the disease was being properly managed. For instance, carers reminded patients to take medication consistently, or to order repeat prescriptions. Some patients relied on (in)formal carers to recognise symptoms of exacerbations or complications, and for medication reminders or assistance using CDM aids.

> *I also have to inject [insulin] every day. … I do it myself. Because I have a device and then I can see the results. And then I tell them, and they [carers] send it to [PN].* (P13)

As patients viewed carers as accessible actors, they trusted their carers with care coordination and information transfer before, during, and/or after consultations. Patients sometimes prepared questions with their carers prior to and during GP consultations. For consultations that are deemed to be important, carers could translate information into more accessible language. The emotional and social support that carers provided to patients helped them to attend appointments.

> *I once went alone [to the GP] and that went okay. But it's nice if someone goes with me to the specialist. … A little support. I also went alone a couple of times and then I cancelled the appointment. … I didn't feel like going alone.* (P11)

HCPs

HCPs indicated that they relied heavily on carers to fulfil the mediator role, supplying them with information on patients' health complaints, (re-)explaining medical information in (more) comprehensible language, and helping to make a treatment plan. They therefore preferred carers to accompany patients during chronic disease consultations.

However, carers' presence during consultations also posed challenges. Firstly, HCPs sometimes had to avoid engaging and making appointments exclusively with carers, who were deemed more efficient, rather than with patients themselves. Secondly, collaborating became more complex when carers were not medically trained, complicating information provision.

> *If a carer comes along, then I hope that they're people with some knowledge of chronic care. … I think they sometimes don't want us to know that they have no idea what you're talking about. So then you're talking to two people who don't really understand, and that's very difficult.* (PN4, FG7)

Despite awareness of patients' networks supporting them with CDM in daily life, the lack of insight into actors and their responsibilities complicated care provision. GPs and PNs had different ways of handling this. Whereas PNs, lacking insights into support networks and with a longer consultation time, undertook tasks that strictly did not belong in their consultation, some GPs ideally functioned as coordinators in support networks for patients with IDs. As patients were limited in their capacity to self-manage their disease properly, this coordinating or referral role was deemed essential. However, GPs often lacked time and resources for extensive support, leaving them with feelings of frustration.

4. Directive decision-making processes

Although most patients mentioned that they expected HCPs making decisions for them, this approach was contradictory to HCPs' preferences. Both groups were aware of medical hierarchy and its influence on decision-making processes.

Patients

Patients valued HCPs' decisions and advice based on hierarchy: GPs were considered more knowledgeable than PNs, but less knowledgeable than medical specialists. However, appointments with highly knowledgeable HCPs for disease monitoring could

cause more distress. In addition to hierarchy, comprehensible information provision and trusting relationship also affected patients' view of HCPs' knowledgeability.

Although patients wanted to be included in the decision-making process, most patients expected that HCPs would ultimately make the final decision for them, especially when they perceived HCPs as highly knowledgeable.

> *If there are abnormalities that we have questions about, then she [cardiologist] includes me, like 'these peaks are too high, you can do something with that'. So it's nice that she includes me in everything that's going to happen.* (P9)

Patients were more likely to follow advice when they perceived their HCP as knowledgeable, because they understood better the benefits of doing so.

HCPs

PNs seemed aware of their lower subjective status relative to GPs, as some experienced difficulties with patients valuing their decisions just as much as GPs' decisions. Therefore, they put extra effort into building trust. Both PNs and GPs mentioned facing difficulties in exchanging information effectively during decision-making processes, including medical content and language matching patients' cognitive abilities. PNs also mentioned that usual approaches, such as motivational interviewing, were not applicable to patients with ID. To address these difficulties, some HCPs used the teach-back method to confirm patients' understanding. Many PNs also mentioned using visual tools – mostly pictures on how to manage diseases, such as a person injecting insulin – as their tasks more often entailed conveying practical information.

Exchanging information was deemed difficult: sometimes, HCPs realised that they had overestimated patients' abilities, even though they were aware of patients' ID, and sometimes HCPs assumed beforehand that they were unable to provide clear information, or patients were unable to understand information.

> *I don't have the illusion that I will do everything [right] in one go. … You incorporate those sorts of things immediately: you incorporate failure.* (GP4, FG5)

> *It's also difficult at times to judge how a person will respond. If you're too strict, they don't come back. And if you're not strict enough, then they just come because it's enjoyable.* (PN4, FG7)

Although contrary to their usual approach, most HCPs often applied a more paternalistic approach than with other patients, which they deemed necessary to provide (directive) decisions. They indicated that they provided only information that they considered most important or practical, resulting in limited shared decision-making. Others would wait until patients experienced an exacerbation, using it as leverage to encourage adherence.

Most HCPs also set smaller self-management goals for patients with ID. Although these goals did not inherently differ (e.g., maintaining a healthy lifestyle), they were often less all-encompassing, making them more achievable (e.g., taking the stairs instead of the lift). However, this approach could impact HCPs' own motivation negatively, as progress was slower than with patients without ID.

> *I actually also need small successes [with the patient] and that's not always feasible and I find that difficult. … Yes, a good conversation can also be a small success, something like a joke or whatever, but preferably also small steps related to medicine. I find it difficult when things actually stay the same or get worse.* (GP2, FG4)

5. Support network to assist in achieving and maintaining a healthy lifestyle

Patients and HCPs acknowledged patients' dilemma around independence versus support needed in achieving and maintaining healthy habits.

Patients

Balancing this dilemma could be difficult: despite patients' aspiration for independence, patients acknowledged that they required sufficient internal motivation and carers' stimulus to make healthy choices and to resist unhealthy temptations.

> *Actually, I'd rather not go [dietician]. My carer says: 'just go, just do it because it is important for you'. … In hindsight, I kind of agree with them. I do need a little push.* (P11)

Patients' living context could either facilitate or hinder healthy decisions. Unhealthy meals or shortage of staff to aid in healthy choices limited patients' ability to achieve and maintain a healthy lifestyle.

Most patients perceived small lifestyle modifications as great accomplishments (i.e., eating fewer unhealthy snacks), although not having access to facilities could hamper healthy living. For many patients, essential facilities aimed specifically at people with (intellectual) disabilities were not always accessible, either financially or practically:

> *[Swimming] is the only sport I can still do because of my back. … Yes, everything in life is getting more expensive. And there are funds where you can get money from your municipality to do recreational things, but still … I [also] have to pay a taxi with that money …. It's not always easy, but I try.* (P12)

HCPs

Despite patients' small lifestyle achievements, HCPs often expected or wanted bigger changes, leaving them with a dilemma on how to address lifestyle. Some mentioned that they were cautious, as patients often already faced multiple problems in different life domains simultaneously. Some GPs dealt with this hesitation by not discussing lifestyle at all or by being more lenient with protocols for discussing lifestyle with patients with ID than for patients without ID.

> *Obviously, you're supposed to do and ask and examine a number of things [at a consultation]. And you can do the examination while you chat. … And while you're talking, you often hear information about all of the things you have to tick off.* (PN4, FG7)

HCPs that did address lifestyle mobilised existing support networks as much as possible to attain small goals, which could aid in providing incentives and motivation to keep agreements.

6. Accessible medical information

Both patients and HCPs expressed their preference for accessible medical information, as it would benefit continuity of care.

Patients

Some patients found it frustrating to repeat their medical history, especially when there was no continuity of care:

> *I've seen three cardiologists in two years, never the same one. … Then you have to tell the whole story again. And I say, people, you have the file, just open it. You get tired of that sometimes and then I think to myself, I'll tell my sister that I'm going home because I can't stand it here anymore.* (P6)

Patients with sufficient digital skills found it useful to check online medical files for arrangements and results of clinical examinations, or asked questions online rather than visiting or calling the general practice.

HCPs

HCPs from different care organisations were not always aware of the type and frequency of medication prescriptions and clinical examinations, or whether a patient had a (suspected) ID, because medical records from different care organisations were not always linkable. This complicated care provision for them, as it was complex to determine the content of treatment plans for chronic diseases.

> *We had about three different medication lists for each patient. The pharmacy had one thing, I had something else, and the ID physician had something else again. Just to show that it's far from being integrated, which is also a point, of course, and then we make mistakes.* (GP4, FG6)

DISCUSSION

This qualitative study explored needs of patients with ID and their HCPs, identifying six care needs for suitable CDM. Several overarching observations can be uncovered across these care needs: the importance of a trusting relationship; difficulties recognising ID; expectations on approaches; and the nature of patients' support networks in effective CDM.

First, we found that a trusting relationship between patients and HCPs underpinned suitable care. In the literature, the benefits of a trusting relationship between patients and HCPs are seen mainly in HCPs being more watchful for patients' care needs [30, 31] and simultaneously allowing patients to discuss their health problems more easily [32, 33]. For patients with a chronic disease [34, 35], including patients with ID [32, 36, 37], long-term trusting relationships have been reported to improve health outcomes, healthcare use and effective care provision. For patients with both diagnoses – a chronic disease and an ID – continuity of care is thus even more essential. As people with ID often had previous care experiences of miscommunication, unclarities regarding treatment, and feelings of not being taken seriously [38], building trust required continuity of care and time [39-42]. For this to occur, patients and HCPs mentioned to require at least accessible medical information and sufficient consultation time. As patients mentioned, separate medical records that do not communicate across different care organisations means that they often have to repeatedly share medical history and disclose their ID to different HCPs. This impedes the building of a trusting relationship with HCPs. Regarding the aspect of time, as patients in our study equated a trusting relationship with use of simple language, consultation time also allowed HCPs to

adapt their approach accordingly. Recent pilots in the Netherlands with increased consultation times have shown promising results in terms of quality of care and satisfaction with care from the perspective of both patients and HCPs [43].

Second, because patients inherently linked trusting relationships with adapted communication, recognising ID was essential for establishing a safe environment and meeting specific care needs. During our data collection, several GP focus groups concentrated largely on recognising ID, before enabling them to reflect on CDM in this patient population. Some GP participants mentioned that sharing experiences with other GPs about patients with ID was already eye-opening, displaying their lack of knowledge and experience with patients with ID. HCPs' lack of knowledge on patients with ID and concomitant communication difficulties are well documented [33, 36, 40, 44-46]. Additional training on ID, for both HCPs and current medical students, as mentioned in several focus groups, is thus essential for effective information transfer and ID recognition [47].

Third, HCPs faced difficulties with suitable communication approaches in patients' CDM, because of patients' dilemma around autonomy versus requiring support patients accepted directive decision-making but also appreciated being involved in the process. As they experienced the inappropriateness of approaches such as shared decision-making or motivational interviewing for patients with ID, HCPs sometimes resorted to more paternalistic approaches, thereby being more steering and directive during decision-making processes. However, previous studies show that more paternalistic approaches potentially deprive patients of their capacity to engage in self-direction over their chronic disease [48]. The literature highlights the potential of modified approaches like supported decision-making or modified motivational interviewing, as they can be useful for managing and meeting patients' expectations, without necessarily adopting a paternalistic approach [49, 50]. These approaches lead towards a more person-centred approach, which can be beneficial for patients with complex care needs, because, like the patients in this study also mentioned, it may increase patients' motivation for treatment adherence and maintaining a healthy lifestyle [49, 51]. Future research can explore how HCPs can be supported in decision-making processes with these patients.

Fourth, despite the importance of patients' support network in their CDM, HCPs often lacked a comprehensive overview of (responsibilities of) involved actors. As HCPs mentioned in our study, exploring these networks resulted in additional workload, as they had to invest more time in finding out which actors carried which responsibility. Similar to previous qualitative studies, we found that even for actors within these networks, their responsibilities are crucial but often unclear [52, 53]. In one study,

multiple actors would take responsibility over a small part of patients' health, resulting in a lack of overview and overall responsibility [52]. Next to providing care and support, involved actors therefore also required additional skills to take responsibility and to adequately support patients before, during, and after CDM consultations [53].

Strengths and limitations

As only the first two focus groups were observed by an assistant, some non-verbal cues could have been missed in the other focus groups. However, the similarity between the moderator's and the assistant's fieldnotes in the first two focus groups and logistical reasons (planning and timing of the focus groups) made us choose to perform the other focus groups without an assistant.

Acquaintance among GPs within the focus groups may have influenced the discussion. Although familiarity can facilitate the disclosure of sensitive topics [54], it may also induce socially desirable answers [55]. Nevertheless, an attempt was made to ensure a safe environment in at least two ways. It was explicitly stated that anonymity was guaranteed and that there were no right or wrong answers. Additionally, each group discussion included only one type of HCP (GPs or PNs). This way, the naturally occurring medical hierarchy within general practice that could possibly impact feelings of safety was eliminated [56]. Consequently, the participating HCPs were very reflective and open about their (in)experiences with people with ID.

Additionally, we included a diverse group of HCPs in terms of age, sex, location, and affinity/experience with people with ID. Although the majority of GPs were located in one region of the Netherlands, the online focus groups allowed us to include a more diversely located group of HCPs. Despite recruiting PNs via different channels, the PNs who participated in the (online) group discussion all had relatively much affinity and knowledge about people with ID. Therefore, the views of the PNs included in this study might differ from those of PNs with less experience with people with ID.

By including perspectives of both patients and their main HCPs, we were able to explore patients' care needs, HCPs' needs in providing adequate CDM, and similarities and differences in these groups' perspectives. Although we included a rather diverse group of patients, in terms of age, sex, living arrangement, and location of residence, most interviewees (n=10/14) worked as 'experience experts' because of difficulties in reaching chronically ill patients with ID. For this job, they had received training in communication and reflection. As this allowed them more than people without such training to voice their experiences, our findings are possibly more in-depth than otherwise.

Implications

Because we used the Chronic Care Model, the outcomes of this study can be interpreted in the light of CDM in Dutch general practice. Although this model contributes to patients being informed and activated, and HCPs being proactive and prepared [57], patients with ID require modified communication approaches and CDM goals to achieve this. With this model as the underlying framework, we found that involving carers in agreements and information transfer is crucial for CDM. It is recommended to plan increased consultation time, as this allows HCPs to develop trusting relationships with both carers and patients and to adapt communication strategies accordingly [7, 41, 58], allowing for continuity of care.

Openly discussing expectations about the CDM process and content can enhance information transfer and avoid feelings of misunderstanding [59]. For GPs, access to, and knowledge of, easy-language information, websites, and visual tools on chronic diseases (e.g., Steffie.nl, Thuisarts.nl) is essential to facilitate patients' understanding of the necessity of CDM.

Given to the importance of patients' support networks in effective CDM, future research is encouraged to include their perspectives also. In particular, carers who assist patients with information transfer during consultations and with CDM within the home situation could provide valuable perspectives to gain a more complete overview of support surrounding patients' care needs.

This study amplified the need for more attention on effective CDM for people with ID. Chronic disease guidelines and CDM protocols should incorporate information on necessary modifications for suitable CDM in vulnerable patient populations, like those with ID, such as communication approaches, goal setting, and activating support networks in CDM. Incorporating ID in GP and PN training, as well as access to information or guidelines on approaching patients with ID in general practice, may enhance effective communication [36, 44, 45, 60] and thereby CDM quality.

Conclusions

This study explored patients' and HCPs' needs in the context of CDM in general practice. Patients required a trusting relationship with their HCP, clear expectations, support in CDM and healthy lifestyle, directive decision-making, and accessible medical information. HCPs largely recognised these care needs, but organisational factors and lack of training or experience with patients with ID hampered the full adjustment of CDM to these needs. More attention in research, policy, and clinical practice is necessary to stimulate the suitability of CDM for patients with ID.

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APPENDIX

Supplementary File 1: Interview guide
Based on domains of Chronic Care Model:

Introduction:
What chronic disease(s) do you have?

Continuity of care/organisation of care:
What doctor do you have for your disease? What do you think about them?
Can you tell me what usually happens if you have to visit your doctor for a check-up for your chronic disease?

Decision support:
If you have a question about your disease, to whom do you go? Why that person?
What information did your doctor gave you when you were diagnosed?

Self-management:
What do you have to do at home for your chronic disease? Do you get help with that?
What is (not) going well?

Supplementary File 2: Focus group guide
Part 0. Introduction
- Short introduction to focus group structure
- Participants introduce themselves: name, age, profession, experience with ID
- Definition of ID and how healthcare providers (HCP) might notice.

Part 1. On ID in general:
- Short discussion of cartoon (see below) of a patient and an HCP: take a specific patient with (suspected) ID in mind, or a typical patient with ID, and fill in the speech bubbles.
- Discuss and use as conversation opener on ID in general.

Part 2. On chronic disease management:

- To what extent are there any differences in providing chronic disease management to patients with or without an ID?
- How do you ensure that you transfer (chronic disease) information in a suitable way to a patient with ID?
- What need(s) do you have in providing chronic disease management to patients with ID?

Part 3. Care needs/wishes:

- What would you wish for your chronically ill patient with ID regarding the chronic disease management that they receive?
- What would you wish for other GPs in providing chronic disease management to patients with ID?

GENERAL DISCUSSION

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This thesis explored the suitability of chronic disease management in general practice for patients with ID. Combining all insights, we propose that general practice holds promise for these patients, especially when meeting three preconditions that could improve suitability of care. The chapter ends by discussing the strengths and limitations of this thesis and the implications for research, policy, and clinical practice.

INTERPROFESSIONAL PERSON-CENTRED APPROACH

Although people with and without ID receive similar amounts of chronic disease management within disease management programmes (Chapter 5), comorbidities are still more prevalent in chronically ill patients with ID than in those without ID (Chapter 3). This implies that their chronic disease management is more complex, but possibly not fully meeting their needs (Chapter 6). In other words, the findings of this thesis point towards *health equality* between people with and without ID in their chronic disease management, but not *health equity*. Thus, despite the similar frequencies with which patients with and without ID receive chronic disease management, the more complex care needs of patients with ID may indicate a need for a different approach for people with ID than for those without ID. By adopting an interprofessional person-centred approach, more health equity could be achieved, eventually leading to more suitable chronic disease management for patients with ID.

Person-centred care, also referred to as personalised or individualised care, prioritises patients' preferences, beliefs, and contexts including families and carers, and places the individual, rather than their symptoms or disease, at the centre of care [1-3]. In a person-centred approach, instead of patients being passive recipients of medical interventions, they participate actively in their care and decision-making processes [4]. Interprofessional collaboration entails collaboration between patients, healthcare professionals, caregivers, and other actors in patients' support networks towards improving patients' health [5]. By addressing a problem with multiple disciplines (e.g., GPs, physiotherapists, behavioural scientists, and psychologists) going beyond their individual disciplinary boundaries [6], more tailored suitable care provision for people with ID can be achieved. Such collaborative effort is vital for addressing their increased risk of chronic diseases, increased prevalence of comorbidities, and more complex care needs (Chapters 2, 6).

In contrast, the one-size-fits-all approach of disease management programmes, that provide highly protocolised care for a single chronic disease, can hamper consideration of patients' unique care needs. Although these programmes have improved coordination between healthcare professionals, these professionals are not fully motivated to collaborate with stakeholders outside of primary care, such as social work or medical specialists, limiting the scope of interprofessional collaboration [7]. Furthermore, disease management programmes are designed to manage solely medical aspects of single diseases, rather than taking a holistic approach, considering the social context [8]. This standardised approach is therefore not always adequate for addressing the increasing complexity of patients' needs [9].

To address these critiques, pilots are currently taking place to move towards a harmonisation of person-centredness and high-quality chronic disease management [e.g., 7, 8, 10, 11]. This shift is promising for patients with ID, for whom recognition of their complex care needs is essential for high-quality chronic disease management. Additionally, embracing person-centred care can potentially reduce the already high healthcare costs of patients with ID [12, 13]. To promote person-centred care, general practice occupational association InEen has outlined a framework emphasising themes deemed important for a valuable transition: positive health, shared decision making, and collaboration [14]. Positive health is a holistic view on health, in which a person's possibilities rather than their ill-health is central [15], for which dialogue tools can aid healthcare professionals identify these possibilities. However, both these dialogue tools and shared decision making (Chapter 6) require some adaptations if they are to be used in patients with ID.

Despite existing initiatives, at least three preconditions should be met for patients with ID to achieve more suitable chronic disease management in the form of a more interprofessional person-centred approach: a supportive policy environment, supportive guidelines, and incentives for providing high-quality care.

There should be a supportive policy environment for interprofessional person-centred care

Policy frameworks should be designed in such a way that they support interprofessional person-centred care. Recent policy frameworks at local, regional, and national level seem promising: the Integral Care Agreement, the national Prevention Agreement, and the Health Policy Memorandum all revolve around improving suitability of care by preventing unhealthy lifestyles, emphasising persons over patients, and stimulating interprofessional approaches [16-18].

Although the policy environment seems supportive of interprofessional person-centred care, the underlying principles of suitable care [19] as stipulated in the Integral Care Agreement require some adjustments by stakeholders in order to achieve (more) suitable chronic disease management for patients with ID. These principles form the collective direction for stakeholders working on suitable care [19]. They emphasise that care should involve patients in care decisions, should be value-driven, deliver the right care in the right place, and shift the focus from disease to health. The former two in particular require some attention to be applied adequately to patients with ID also.

The principle of care being developed with and around patients, considering their abilities, highlights the necessity of suitable decision-making processes for patients with ID. Given their limited health literacy [20], decision-making processes should occur with the involvement of carers to enable patients' participation in healthcare decisions (i.e., *supported* decision making; not to be confused with *substituted decision making*). (In) formal carers then function as mediators between healthcare providers and patients (Chapter 6). For healthcare providers and carers, knowledge on how to support patients adequately is essential for collaboratively developing care. Inherently, for people with ID, this principle emphasises the significance of collaboration between primary healthcare providers and professionals from the social domain who provide support to patients with ID. Knowledge of patients' support networks is thus important (Chapter 6).

Additionally, value-driven care, meaning that it is effective care based on science and practice, is essential. As shown in this thesis (Chapter 2), studies on people with ID often involve small, non-representative groups, have varying definitions of ID, and use different methods for identifying people with ID. Without adequate (scientific) knowledge, the step towards suitable care cannot be made, as guidelines do not support healthcare providers sufficiently. This is elaborated upon below.

There should be supportive guidelines for adequately managing chronic diseases
Guidelines are important in improving and maintaining quality of care, as they contain recommendations based on expertise and experiences of healthcare providers and healthcare users, scientific evidence, and considerations of the (dis)advantages of treatment options [21, 22]. However, current guidelines issued by the Dutch College of General Practitioners (Dutch: NHG) are often based on the average patient [21], excluding more complex patient populations such as those with ID.

The absence of ID in primary care guidelines is mirrored by the lack of acknowledgment of ID in the NHG's research priorities. The stakeholder-driven research agenda tends to overlook patients with ID, even with the participation of

HandicapNL [23, 24]. Also, summaries on missing scientific evidence for clinical recommendations (i.e., the NHG's knowledge gaps [25]) do not include stratification between patient populations, such as those with and without ID. Consequently, healthcare providers are left without the necessary support to make informed decisions, resulting in either adherence to possibly unsuitable guidelines or non-compliance based on personal experience. Supporting healthcare providers with relevant information is thus essential for suitable chronic disease management.

Suitability of care can be increased by adding ID to existing guidelines. Both disease-specific guidelines (e.g., management of type 2 diabetes mellitus) and multidisciplinary guidelines provide opportunities for including ID. In disease-specific guidelines, addendums should be added in which the different approaches and health patterns of people with ID are described. Additionally, the existing multidisciplinary guideline on identifying pain and physical health problems in people with ID should expand its scope to recognising ID and chronic disease management.

There should be incentives to provide high-quality chronic disease management
Healthcare providers should be incentivised to maintain a high quality of chronic disease management with an interprofessional person-centred approach, especially for patients with ID, who are at increased risk of chronic diseases (Chapter 3). Specifically those who are not enrolled in disease management programmes, and for whom registration of quality indicators is not optimal (Chapters 4, 5), should receive high(er) quality chronic disease management. Several possibilities are proposed in the literature.

First, sufficient consultation time is crucial. A recent pilot showed that, when GPs had more consultation time, they prescribed less medication, experienced more effective time per patient, and had a lower workload [26]. Albeit seemingly promising, increased consultation time alone is not sufficient for suitable care for patients with ID. As shown in Chapter 6, healthcare providers should also adapt their communication skills, set smaller disease management goals, and involve carers in agreements. Thus, healthcare providers require additional skills and competencies to recognise patients' needs and to provide person-centred care [27]. The involvement of practice nurses, who often have more consultation time than GPs, can benefit the patient–provider relationship and build trust (Chapter 6).

Second, individual care plans in patients' chronic disease management can be beneficial for the quality of care experienced [28]. These plans are developed with patients and healthcare professionals and are translations of care standards to

individual goals and agreements with patients [29]. They go beyond disease-specific measures (e.g., HbA1c) by considering patients' context [7]. For patients with ID, it has been found that individual care plans can aid in improving self-management skills, community involvement [30, 31], and reducing stigmas on their diagnostic label [32]. As these plans should be tailored to patients' needs, it is important that they are 'living', easy-language documents, in which information is broken down into smaller pieces [32]. Furthermore, both *care planning* (collaboratively discussing and agreeing on goals) and *care plans* (written documents containing the outcomes of a care planning process) are needed [33] to safeguard the quality of chronic disease management.

Third, the national primary care collaboration agreement (LESA) for providing care to frail elderly patients is a promising incentive to stimulate interprofessional collaboration [34]. This agreement contains descriptions of divisions of relevant stakeholders' tasks and responsibilities, prioritising patients' needs and quality of life. Although the shortage of ID physicians and their relative unfamiliarity to other healthcare providers [35, 36] complicates collaboration, more suitability of care can be achieved through clear communication and concrete agreements between GPs and ID physicians, especially in long-term contexts such as chronic disease management. The person-centred and interprofessional nature of LESAs can aid in addressing (and possibly overcoming) complexities in care provision for vulnerable patient populations, such as patients with ID.

STRENGTHS AND LIMITATIONS

To the best of our knowledge, this thesis is the first to examine the suitability of chronic disease management for patients with ID. To better interpret information and to gain a rich picture on chronic disease management for people with ID, we applied both quantitative and qualitative research methods. All individual studies were closely intertwined, with each study building upon the findings of the previous one.

The research team and advisory board consisted of people from different backgrounds, adding to a broad interpretation of findings. The research team was composed of health researchers and physician researchers; the advisory board consisted of healthcare providers from various backgrounds, working in various organisations and regions. When necessary, additional external advisors (e.g., statisticians, co-researchers with ID) were consulted to improve the quality of research.

We provided a multifaceted perspective and rich understanding of chronic disease management for people with ID by presenting information on a heterogeneous and large group of people with ID and by including perspectives of both GPs and practice nurses. By utilising large-scale datasets, we were able to explore a more representative and heterogeneous group of chronically ill people with ID (ranging from suspected to severe IDs, and from residing in community to being in residential care) than by solely using smaller-scale data and/or convenience samples. Additionally, because of the important role of practice nurses in chronic disease management in the Netherlands, we also included their views. Due to its broad perspective, this thesis is therefore also likely to have implications for a more extensive patient population, particularly those with lower health literacy.

Despite being able to include a rather heterogeneous group of people with ID, we were yet unable to stratify results from our studies by ID aetiology or severity. For most patients, no such information was available. As ID aetiology is often multifactorial, caused prenatally, perinatally, or postnatally by biomedical, behavioural, social, and/or educational factors [37], different causes may have different implications for health. Specific syndromes may have increased risk of chronic diseases, such as the increased risk of cardiovascular disease for people with Down syndrome [38]. Thanks to the explorative nature of our studies, this thesis provided a first glance at chronic disease patterns in people with ID. Future research is encouraged to include aetiology in studies of (risk for) chronic diseases.

Additionally, despite difficulties distinguishing asthma and COPD in people with ID [39, 40], in Chapter 6 we included both diagnoses, whereas in Chapters 2–5 we considered only COPD. By including only COPD in our quantitative studies, we were able to study patients of whom we could say with more certainty that they had a COPD diagnosis, rather than including both asthma and COPD. However, it is likely that, in reality, a larger group of patients would have been eligible for inclusion.

IMPLICATIONS FOR RESEARCH, POLICY, AND CLINICAL PRACTICE

This thesis has important implications for research, policy, and clinical practice that revolve around attention on ID, healthcare providers' skills, and tailoring care.

First, this thesis highlights the importance of more attention being paid to ID in policy, medical education, and medical literature. A brief PubMed search in the top 10 medical journals revealed that a mere 0.12% (1,154 articles) mentioned ID in their

title and/or abstract.[1] This lack of knowledge of this vulnerable patient population is reflected in patients' chronic disease management. Despite extensive research on chronic diseases and ID individually, research on both diagnoses combined remains relatively scarce. People with ID should be included in clinical guidelines by incorporating specific information about ID into existing (chronic disease) guidelines or through a separate guideline on ID.

Second, healthcare providers' skills should be improved. Given the importance of approaching patients in a manner suitable to their needs and abilities, increasing healthcare providers' exposure to patients with ID is likely to increase their competence and comfort in care provision [41]. It could therefore be considered whether a mandatory number of hours dedicated to vulnerable patient populations should be integrated into medical school curricula, similar to existing mandatory training in communication skills for collaboration in some multidisciplinary teams [42]. Given the added value of practice nurses in patients' chronic disease management in terms of continuity of care and relieving GPs' workload [43], it could be beneficial to offer them (additional) training on approaching ID. A deeper understanding of the communication and healthcare needs of patients with ID is an essential foundation for suitable chronic disease management.

Third, chronic disease management should be better tailored to patients' needs. To do so, increased understanding of their needs is necessary: future research should focus on the possible links between (risk for) chronic diseases and ID aetiology. This could aid in developing targeted interventions and preventive measures to decrease the burden of chronic diseases for patients with ID.

Tailoring disease management to patients' needs also requires increased utilisation of available resources. Despite the existence of such resources for recognising, approaching, and communicating with people with ID, healthcare providers often underutilise them. Useful tools include, for instance, an online tool that guides healthcare providers towards more person-centred communication in primary care settings [44], a tool that aids them to better incorporate the needs of patients with

1 The top 10 journals in the broad Health & Medical Research category according to the Google Scholar journal ranking from 1955 to 14 August 2023. Ranked from first to last place, these journals were: *The New England Journal of Medicine*, *The Lancet*, *Cell*, *JAMA*, *Nature Medicine*, *Proceedings of the National Academy of Sciences*, *BMJ*, *International Journal of Molecular Sciences*, *Journal of Clinical Oncology*, and *PLoS ONE*. In these journals, in PubMed, titles and abstracts were screened for the occurrence of one of the terms intellectual disability/-ies, learning disability/-ies, mental retardation.

limited health literacy in daily care provision [45], and a screening tool to aid in diagnosing diseases at an earlier stage [PROSPER-ID; 46]. Future research should explore underutilisation of resources and strategies to encourage their utilisation.

Additionally, increased collaboration is required to facilitate the preconditions necessary for suitable chronic disease management for patients with ID. Collaboration between the ID sector (e.g., the Dutch association of healthcare providers for people with ID VGN) and primary care (e.g., NHG) and between ID physicians and GPs could aid in tailoring care to patients' needs. Collaboration between ID physicians and GPs could improve diagnostic processes and the suitability of chronic disease management [47, 48]. The development of a LESA specifically targeted at people with ID is recommended to improve interprofessional collaboration between relevant stakeholders. Lastly, within the standardised disease management programmes, possibilities should be considered either to develop such programmes specifically tailored to patients with ID or to incorporate patients with ID in programmes designed for other vulnerable patient populations, such as frail elderly patients.

Overall, more attention on ID in research, policy, and clinical practice is a first step towards awareness of the differences between people with and without ID. Increasing healthcare providers' knowledge of ID, of patients' risk for chronic diseases, and their needs in chronic disease management is a first step towards more suitable chronic disease management. The use of existing resources to expand knowledge should be stimulated. Stakeholders should also be incentivised towards more interprofessional collaboration and person-centred approaches. Eventually, these will pave the way towards more health equity for patients with ID.

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SUMMARIES

ENGLISH SUMMARY

Because of the high impact of chronic diseases such as cardiovascular disease, diabetes mellitus type 2, and chronic obstructive pulmonary disease (COPD), it is essential that chronic disease management is of high quality and is suitable for patients with intellectual disabilities (ID). Around 1.5% of the Dutch population is estimated to have an ID, although an additional 6.4% of the population also has characteristics of ID. People with ID are characterised by extensive limitations in intellectual functioning and adaptive behaviour, which may complicate understanding and communicating (symptoms) of diseases. Despite these differences with the general population, people with ID receive chronic disease management in regular general practice. However, it is unknown yet to what extent their disease management is suitable to their needs. This thesis therefore described the prevalence of chronic diseases, care provision, and care needs in people with ID, with the overall aim to examine the suitability of chronic disease management in general practice for this patient population.

We explored the different prevalence rates of chronic diseases in the literature in **Chapter 2**. With a scoping literature review, we mapped the broadness of published literature on chronic disease prevalence, and explored main characteristics of included studies. We found that in the 19 studies that were included in our final sample, the prevalence of chronic diseases varied considerably between people with and without intellectual disabilities. Especially the type of data, method of identification of ID, country in which the study was performed, age groups taken into account, and sample size appeared important in these differences. Higher prevalence of cardiovascular disease was found among studies using support as proxy for ID-diagnosis. Also, in the USA, prevalence rates of chronic diseases were generally higher than in other countries. Age appeared important, as studies focusing on all ages reported the lowest prevalence rates of chronic diseases. In studies with larger sample sizes prevalence rates in general were lower than in studies with higher sample sizes. We therefore suggested that primary care providers and researchers should interpret results on disease prevalence among people with ID in the context of methodology and in light of the study characteristics.

In **Chapter 3** we described our findings on prevalence of cardiovascular disease (ischaemic heart disease and cerebrovascular disease), diabetes mellitus, and COPD, as well as comorbidity patterns among people with ID compared to people without ID. We combined data from Nivel Primary Care Database with population data from Statistics Netherlands to compare people with and without ID. At group level, 14.9% of people with ID and 16.9% of people without ID had a diagnosis of at least one chronic disease. When

considering age and sex, cerebrovascular disease (prevalence rate (PR)=1.1), diabetes mellitus (PR=1.6), and COPD (PR=1.5) were more prevalent in people with than without ID, and ischaemic heart disease (PR=0.7) was less prevalent. At younger age, people with ID also more often had a chronic disease and more often had additional comorbidities than people without ID. Although in the general population women had the highest disease burden in terms of prevalence of chronic diseases and comorbidities, in the group of people with ID males most often had a chronic disease and multiple comorbidities. Most often comorbidities were of circulatory nature. Even with probable underdiagnosis of chronic diseases due to difficulties recognising symptoms of disease in people with ID, these results suggest that people with ID are more at risk of chronic diseases than people without ID. Awareness of age and sex differences between people with and without ID are important for timely recognising diseases.

Our findings on recordings of disease monitoring in residential care settings are described in **Chapter 4**. Utilising patient-level medical record data between July 2020 and December 2021 from a large long-term care provider for people with ID, we examined recordings of chronic disease management of 352 patients with ID by exploring consultation rates and recordings of quality indicators. These quality indicators are developed to ensure the quality of chronic disease management and relate to clinical aspects (e.g. blood pressure measurement) and healthy lifestyle (e.g. discussing smoking behaviour). We found that the median number of consultation rates for patients with cardiovascular disease (Median=2.0), for patients with type 2 diabetes mellitus (Median=4.7), and for patients with COPD (Median=2.7) was consistently lower than the baseline number of consultations in general practice. Few patients with cardiovascular disease, type 2 diabetes mellitus, or COPD had a complete profile of lifestyle and disease-specific quality indicators. Especially discussion of lifestyle was poorly recorded. Disease-specific quality indicators were recorded more often in patients' medical files, but at low frequency. For patients with cardiovascular disease and type 2 diabetes mellitus, a record of an eGFR test was most often occurring in their medical files. Overall, recording of quality indicators was most often incomplete for patients with COPD. The recording of chronic disease management for patients with ID in residential care settings appears suboptimal. This may have implications for quality of care, as it limits opportunities for evaluation and improvement of chronic disease management.

Chapter 5 addressed differences between patients with and without ID in the enrolment in disease management programs and in disease monitoring in general practice. By combining data from Nivel Primary Care Database with population data from Statistics Netherlands, we could match 2,653 patients with ID and a chronic disease to 13,265

patients without ID (1:5). With conditional logistic regression analysis we found that independent of enrolment in a disease management program, patients with ID were more likely than those without ID to frequently consult their general practitioner. There were few differences in enrolment in disease management programs between those with and without ID: patients with ID were only more often enrolled in the diabetes management program, while there were no differences in enrolment in the cardiovascular or COPD programs. Within disease management programs, patients with and without ID received medication prescriptions and routine examinations in similar frequencies. Among those not enrolled in the cardiovascular or COPD program, patients with ID more often received medication prescriptions and routine examinations than those without ID. Despite previous studies reporting care inequities between people with and without ID, we did not find evidence for limited access to disease management programs for patients with ID and a chronic disease.

In **Chapter 6**, we reported our findings from 14 interviews with patients with ID and seven focus groups with healthcare providers on their care needs in the context of chronic disease management. We identified several care needs underpinning to suitable chronic disease management. A trusting relationship between patients and healthcare provider was an important precondition for suitable disease management. Clear expectations about the process of chronic disease management, and accessible medical information was discussed as being helpful in chronic disease management. Patients' support networks played vital roles in managing their chronic diseases and in achieving and maintaining a healthy lifestyle. Contrary to patients without ID, healthcare providers noticed patients' preference for more directive decision-making processes, while patients also appreciated some level of involvement in the process. As patients with ID often required different approaches than patients without ID (such as supported decision making, or modified motivational interviewing), adapting communication styles was necessary for information transfer. Organisational factors and lack of training or experience could hamper healthcare providers to meet these needs.

The final chapter, **Chapter 7**, contains a reflection on the main findings of this thesis and presents several preconditions for (more) suitable chronic disease management for patients with ID. Additionally, the strengths, limitations, and recommendations for clinical practice, policy, and research are discussed.

NEDERLANDSE SAMENVATTING

Vanwege de aanzienlijke impact van chronische ziekten zoals cardiovasculaire ziekte, diabetes mellitus type 2, en chronisch obstructieve longziekte (COPD), is het van belang dat chronisch ziektemanagement van hoge kwaliteit is en passend is voor mensen met een verstandelijke beperking (VB). Het wordt geschat dat ongeveer 1.5% van de Nederlandse bevolking een VB heeft, alhoewel nog eens 6.4% van de bevolking ook kenmerken van VB heeft maar (nog) geen diagnose. Mensen met VB worden gekenmerkt door significante beperkingen in het intellectueel functioneren en in het adaptief gedrag, waardoor het begrijpen en communiceren van (symptomen van) ziekte moeilijker is. Ondanks deze verschillen met de algemene bevolking ontvangen mensen met VB zorg voor hun chronische ziekte veelal in de reguliere huisartsenpraktijk. Het is echter nog onbekend in hoeverre deze zorg passend is bij hun behoeften. Dit proefschrift beschrijft daarom de prevalentie van chronische ziekten, chronisch ziektemanagement, en zorgbehoeften bij mensen met VB, met als overkoepelend doel het onderzoeken van de geschiktheid van chronisch ziektemanagement in de huisartsenpraktijk voor deze patiëntpopulatie.

In **Hoofdstuk 2** verkenden we de verschillende prevalentiecijfers van chronische ziekten in de literatuur. Middels een *scoping* literatuurreview hebben we de breedte van gepubliceerde literatuur over prevalentie van chronische ziekten in kaart gebracht, en kenmerken van geïncludeerde studies onderzocht. We vonden dat in de prevalentie van chronische ziekten bij mensen met en zonder VB aanzienlijk varieerde in de 19 studies die we includeerden in de analyse. Voornamelijk het type data dat werd gebruikt, methode om VB te identificeren, land waarin de studie werd uitgevoerd, de leeftijden die werden aangehouden, en samplegrootte leken belangrijk te zijn bij deze verschillen. Een hogere prevalentie van cardiovasculaire ziekte werd voornamelijk gevonden bij studies die VB-specifieke ondersteuning als indicator gebruikten om mensen met VB te identificeren. Ook bleek dat prevalentiecijfers van chronische ziekten in de Verenigde Staten over het algemeen hoger waren dan in andere landen. Studies die onderzoeksgroepen van alle leeftijden bekeken, rapporteerden de laagste prevalentiecijfers van chronische ziekten vergeleken met studies die onderscheid maakten tussen leeftijdsgroepen. In studies met grotere samplegroottes waren prevalentiecijfers over het algemeen lager dan in studies met kleinere samplegroottes. Voor eerstelijnszorgverleners en onderzoekers is het daarom van belang om prevalentiecijfers te interpreteren met het oog op de studiekenmerken en in de context van methodologie.

In **Hoofdstuk 3** werden bevindingen gerapporteerd over de prevalentie van cardiovasculaire ziekte (ischemische hartziekte en cerebrovasculaire ziekte), diabetes mellitus en COPD, evenals patronen van comorbiditeiten bij mensen met

VB in vergelijking met de algemene bevolking. We combineerden data van Nivel Zorgregistratie Eerste Lijn met populatiedata van het Centraal Bureau voor de Statistiek om zo mensen met en zonder VB te vergelijken. Op het eerste gezicht had 14.9% van de mensen met VB (2,653 mensen) en 16.9% van de mensen zonder VB (184,681 mensen) een diagnose van ten minste één chronische ziekte. Wanneer leeftijd en geslacht in acht werden genomen, kwamen cerebrovasculaire ziekte (prevalentieratio (PR)=1.1), diabetes mellitus (PR=1.6), en COPD (PR=1.5) vaker voor bij mensen met dan zonder VB, en ischemische hartziekte kwam minder vaak voor (PR=0.7). Al op jongere leeftijd hadden mensen met VB vaker een chronische ziekte en vaker bijkomende comorbiditeiten. Hoewel in de algemene bevolking vrouwen de hoogste ziektelast hadden wat betreft prevalentie van chronische ziekten en comorbiditeiten, hadden mannen binnen de groep met VB het vaakst een chronische ziekte en meerdere comorbiditeiten. Mensen hadden het vaakst hart- en vaatziekten als comorbiditeit. Zelfs met waarschijnlijke onderdiagnose van chronische ziekten bij mensen met VB vanwege moeilijkheden bij het herkennen van ziektesymptomen suggereren deze resultaten dat mensen met VB groter risico hebben op chronische ziekten dan mensen zonder VB. Het is dus belangrijk om zich bewust te zijn van leeftijdsverschillen en verschillen tussen mannen en vrouwen voor het tijdig herkennen van ziekten.

Onze resultaten over het vastleggen van ziektemonitoring in zorginstellingen zijn beschreven in **Hoofdstuk 4**. We hebben gekeken hoe ziektemanagement is vastgelegd in de medische dossiers van 352 patiënten (tussen juli 2020 en december 2021), met de nadruk op aantal consulten en vastleggingen van kwaliteitsindicatoren. Deze indicatoren zijn ontwikkeld om kwaliteit van chronisch ziektemanagement te waarborgen en hebben betrekking op klinische aspecten (bijv. bloeddrukmeting) en op gezonde leefstijl (bijv. stoppen-met-roken-gesprek). Het aantal consulten voor patiënten met cardiovasculaire ziekte (Mediaan=2.0), voor patiënten met type 2 diabetes mellitus (Mediaan=4.7), en voor patiënten met COPD (Mediaan=2.7) waren lager dan de referentieaantallen in de reguliere huisartsenpraktijk. Voor weinig patiënten met VB met (een van) deze drie ziekten waren alle kwaliteitsindicatoren voor leefstijl- en ziektespecifieke tests volledig ingevuld. Vooral het bespreken van leefstijl tussen zorgverlener en patiënt was slecht vastgelegd. Ziektespecifieke kwaliteitsindicatoren waren vaker, maar alsnog weinig, vastgelegd. Voor patiënten met cardiovasculaire ziekten en type 2 diabetes mellitus was het uitvoeren van een eGFR-test het vaakst vastgelegd in het medisch dossier. Voor patiënten met COPD waren kwaliteitsindicatoren over het algemeen het vaakst onvolledig. Het lijkt er dus op dat niet alle kwaliteitsindicatoren worden vastgelegd in de medische dossiers van patiënten met VB wiens chronische ziekte(n) via de zorginstelling wordt gemanaged. Dit kan gevolgen hebben voor de kwaliteit van zorg, omdat de mogelijkheden voor evaluatie en verbetering van ziektemanagement worden beperkt.

Hoofdstuk 5 behandelde verschillen tussen patiënten met en zonder VB in hun deelname aan ketenzorgprogramma's en in ziektemonitoring in de huisartsenpraktijk. Door data uit de Nivel Zorgregistratie Eerste Lijn te combineren met populatiedata van het Centraal Bureau voor de Statistiek konden we 2,653 patiënten met VB en een chronische ziekte koppelen (1:5) aan 13,265 patiënten zonder VB. Met conditionele logistische regressieanalyse vonden we dat patiënten met VB vaker dan gemiddeld op consult kwamen bij de huisarts, ongeacht wel of geen deelname aan ketenzorgprogramma's. We vonden weinig verschillen in deelname aan ketenzorgprogramma's tussen patiënten met en zonder VB. Patiënten met VB waren wel vaker ingeschreven in het diabeteszorgprogramma, maar er waren geen verschillen in deelname aan het cardiovasculair of COPD-programma. Patiënten met en zonder VB die wel deelnamen aan ketenzorgprogramma's werden even vaak medicatie voorgeschreven, en ziekten werden even vaak gemonitord met klinische tests. Voor patiënten die voor hun cardiovasculaire ziekte of COPD buiten ketenzorgprogramma's om zorg ontvingen, kregen degenen met VB vaker medicatie voorgeschreven en werden deze ziekten intenser gemonitord dan patiënten zonder VB. We vonden dus geen bewijs voor beperkte toegang tot ketenzorgprogramma's voor patiënten met VB en een chronische ziekte.

In **Hoofdstuk 6** beschreven we onze bevindingen uit 14 interviews met patiënten met VB en zeven focusgroepen met zorgverleners over hun zorgbehoeften bij chronisch ziektemanagement. Er waren verschillende zorgbehoeften die men essentieel als bestempelde voor passend chronisch ziektemanagement. Een vertrouwensrelatie tussen zorgverlener en patiënt was allereerst een belangrijke voorwaarde voor passend ziektemanagement. Duidelijke verwachtingen over het proces van ziektemanagement en toegankelijke medische informatie werden ook genoemd als helpend voor passende zorg. De (in)formele sociale netwerken van patiënten speelden een cruciale rol bij zowel het chronisch ziektemanagement als het bereiken en behouden van een gezonde leefstijl. In tegenstelling tot patiënten zonder VB merkten zorgverleners dat patiënten met VB behoefte hadden aan meer directieve besluitvormingsprocessen, terwijl patiënten ook betrokken wilden zijn in het besluitvormingsproces. Ten slotte werd toegankelijke medische informatie als essentieel beschouwd voor continuïteit van zorg. Omdat patiënten met VB vaak andere benaderingen nodig hadden dan patiënten zonder VB (zoals *supported decision-making* of aangepaste *motivational interviewing*), was het noodzakelijk om communicatiestijlen aan te passen voor effectieve informatieoverdracht. Organisatorische factoren en gebrek aan training of ervaring konden zorgverleners belemmeren om aan deze behoeften te voldoen.

Het laatste hoofdstuk (**Hoofdstuk 7**) bevat een reflectie op de belangrijkste bevindingen van dit proefschrift. Er worden enkele randvoorwaarden besproken die nodig zijn om passende(re) chronisch ziektemanagement voor patiënten met VB te verkrijgen. Daarnaast worden de sterke punten en limitaties besproken, alsmede aanbevelingen voor de klinische praktijk, beleid, en onderzoek.

DATA MANAGEMENT STATEMENT

DATA MANAGEMENT STATEMENT

Before commencement, initial study protocols for Chapters 3, 5, and 6 were reviewed by the local ethical committee of Radboudumc. For Chapters 3 and 5 the committee waived the need for formal ethical assessment (2017-3921), and ruled for Chapter 6 that this study was not subject to the Medical Research Involving Human Subjects Act (2021-13402). The initial study protocol for Chapter 4 was reviewed and approved by the scientific board of 's Heeren Loo. Study protocols of Chapters 3, 4, and 5 were preregistered at Open Science Framework and made public after an embargo period. All studies in this thesis were conducted in compliance with the principles of the Declaration of Helsinki and the General Data Protection Regulation.

Studies included in this thesis were based on existing literature (Chapter 2), on third-party data (Chapters 3, 4, and 5) and original data (Chapter 6). Data collection and storage was according to the Findable, Accessible, Interoperable and Reusable (FAIR) principles [1]. Explorative analyses for the scoping literature review (Chapter 2) were performed in Excel. These data are stored on the H-station of the Department of Primary and Community Care of the Radboudumc and are available upon reasonable request.

Third-party data underlying for Chapters 3 and 5 were only accessible in the secured environment of Statistics Netherlands. Before publication, Statistics Netherlands performed a check for data adhering to their guidelines regarding anonymity. Therefore, at no time, data was traceable to individuals. Aggregated data are stored on the H-station of the Department of Primary and Community Care of the Radboudumc. They will be archived and stored for 10 years. Datasets from Statistics Netherlands are not publicly available. Aggregated data from the used datasets are publicly available on the website of Statistics Netherlands (http://statline.cbs.nl) and Nivel (https://www.nivel.nl/en/nivel-zorgregistraties-eerste-lijn/nivel-primary-care-database). Non-public microdata are accessible for research purposes, subject to specific conditions and associated fees (see www.cbs.nl).

Third-party data underlying for Chapter 4 could only be accessed by relevant project members. Aggregated data are stored on the H-station and only accessible for relevant team members. They will be archived and stored for 10 years.

Original data was collected with semi-structured interviews and focus groups (Chapter 6). Prior to participation, all participants granted their informed consent. Digital informed consent forms were stored on an external archive where data will be stored

for 15 years. To safeguard the privacy of participants, unique participant codes were utilised, to which only the primary researcher had access. Only relevant members of the project team had access to the anonymised transcripts. Data analyses were performed using Atlas.ti, of which the codebooks with categories and themes are stored in the project file at the department server. As participants did not provide consent to use the data for other purposes than the conducted study, verbatim transcripts are not available.

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1. Wilkinson MD, Dumontier M, Aalbersberg IJ, Appleton G, Axton M, Baak A, et al. The FAIR Guiding Principles for scientific data management and stewardship. Sci Data. 2016;3(1):1-9.


10

DANKWOORD

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Na vier jaar "computeren" is dit mijn laatste stukje schrijven, maar voor velen waarschijnlijk het eerste dat ze lezen. Welkom in mijn proefschrift! Blader vooral nog eens terug naar de onderzoekshoofdstukken, daar zit het meeste werk in. Zoveel lieve mensen om me heen hebben me op een of andere manier gesteund tijdens de totstandkoming van dit proefschrift: collega's, vrienden, en familie. Aan iedereen die zich aangesproken voelt: bedankt voor je steun!

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Met veel plezier heb ik zo'n vier jaar op de 'circustent' gezeten, in de kantoortuin op de derde verdieping, afgebakend met een pot pindakaas en een kopje thee. Met mijn vrijwel dagelijkse aanwezigheid voelde ik me soms toch een beetje onderdeel van het meubilair, maar gelukkig kwamen mijn lieftallige GMVB-collega's vaak samen met mij in 'ons' hoekje zitten. Bedankt voor jullie gezelligheid, ondersteuning, advies, roddels, borrels en uitjes! Ondanks dat ik mijn informele functie als 'hoofd gezelligheid' nu neerleg, zal ik nog eens een verplichte borrel inplannen. :-) Ik draag de circustent graag over aan een waardig opvolger, zorgen jullie er goed voor?

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Natuurlijk ook een blijk van waardering voor alle S.T.A.R. Lads/PhD's: Amina, Anne, Joep, Julia, Katrien, Kim, Manon, Marian, Masha, Meike, Soemeja, Stacey, Wijnand: de weekstart, borrels, etentjes, schrijfclub, 'tour de casa' en promovendi-overleggen waren altijd gezellig en soms zelfs nuttig. Van borrels in de Aesculaaf tot schrijfweken in Maastricht: ik durf wel te zeggen dat mijn PhD minder leuk was geweest als jullie er niet waren. Het was zo fijn om met jullie te sparren en successen (of minder succesvolle dingen) te delen en vieren. Speciale shout-outs naar Katrien en Marian, voor de praktische en mentale steun met de 'proefschrift supportgroep'; paranimf Amina, voor je onmisbare hulp bij mijn "sprookje", de koffiemomentjes, en mentale support; en paranimf Masha voor de gezelligheid, het sparren en delen van ervaringen. Leuk dat we het samen zijn begonnen en het nu (voor mij) ook samen afsluiten!

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te meer. Voor mij was het de ultieme combinatie om naast mijn PhD (waarbij het werk nooit af is) studenten en andere PhD's te coachen en ze met praktische vragen op weg te helpen. Ook voor mijn eigen schrijfproces bleek het best nuttig; regelmatig kwam ik weer aanzetten bij mijn collega's met "hebben jullie wel eens van kernzinnen gehoord?".

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ABOUT THE AUTHOR

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Milou van den Bemd werd geboren op 23 maart 1995. Na het afronden van haar vwo-diploma aan de Katholieke Scholengemeenschap Etten-Leur verhuisde ze naar Nijmegen om de bachelor Sociologie aan de Radboud Universiteit te volgen. Daarna volgden de Master of Research in Sociology & Demography aan de Universitat Pompeu Fabra in Barcelona, Spanje, en de Master of Science in Sociology aan Tilburg University (cum laude). In haar masterthesis onderzocht ze het operationaliseren, testen, en toepassen van de 'American default lifestyle' in de Nederlandse setting. Na bij iresearch als projectmedewerker te hebben gewerkt, begon ze in 2019 aan haar PhD bij het Radboudumc. Naast allerhande onderzoekswerkzaamheden zette ze zich ook in als academisch schrijfcoach, als lid van diverse werkgroepen en de PhD council, en rondde ze succesvol een trainingsprogramma af voor onderzoekers in de eerste lijn (TUTOR-PHC: Transdisciplinary Understanding and Training on Research – Primary Health Care). Milou woont in Nijmegen.

Milou van den Bemd was born on March 23, 1995. After completing her secondary school at Katholieke Scholengemeenschap Etten-Leur, she moved to Nijmegen to pursue a Bachelor's degree in Sociology at Radboud University. She graduated the Master of Research in Sociology & Demography at Universitat Pompeu Fabra in Barcelona, Spain, and the Master of Science in Sociology at Tilburg University (with honours). Her master's thesis focused on operationalising, testing, and applying the 'American default lifestyle' in the Dutch context. After working as a project officer at iresearch, she started her PhD at Radboudumc in 2019. In addition to her research activities, she worked as academic writing coach, was a member of various working groups and of the PhD council, and successfully completed a training program for researchers in primary health care (TUTOR-PHC: Transdisciplinary Understanding and Training on Research – Primary Health Care). Milou lives in Nijmegen.

PHD PORTFOLIO OF MILOU VAN DEN BEMD

Department: **Primary and Community Care**

PhD period: **16-09-2019 – 31-12-2023**

PhD Supervisor: **Prof. dr. G.L. Leusink**

PhD Co-supervisors: **dr. M. Cuypers, dr. B.W.M. Schalk, dr. E.W.M.A. Bischoff**

Training activities	Hours
Courses	
• Workshop PubMed (2019)	2.00
• RIHS - Introduction course for PhD candidates (2019)	15.00
• RU - Digital Tools (2019)	4.00
• Literature review for your PhD (2019)	2.00
• Designing a PhD research project (2020)	84.00
• Medische basiskennis (2019-2020)	224.00
• EBROK (2020)	42.00
• Principles of Research in Medicine and Epidemiology (2020)	15.00
• Training Communicatievaardigheden (2020)	8.00
• Presentatietraining Vanzelf Spreken (2020)	8.00
• Scientific Writing for PhD candidates (2020)	84.00
• Open Science for PhD candidates (2020)	28.00
• Radboudumc - Scientific integrity (2021)	20.00
• Project management for PhD candidates (2021)	56.00
• The Art of Presenting Science (2021)	42.00
• Kwalitatief onderzoek in de gezondheidszorg (2021)	20.00
• Mindfulness-Based Stress Reduction (2021)	28.00
• Cursus focusgroepen modereren (2022)	16.00
• Career workshop: Next step in your career (2022)	15.00
Seminars	
• PhD meeting Intellectual Disability & Health (2019-2023)	80.00
• Opening AI for Health and Thira Lab (2019)	2.00
• Research Round What's wrong with digital health? (2019)	2.00
• Webinar Publishing in Open Access (2020)	1.00

Training activities	Hours
• Webinars (3) Inclusieve wetenschap: omdat iedereen meetelt! (2020)	3.00
• Webinar series on COVID-19 and intellectual disabilities (2020)	2.00
• Research Integrity Rounds: Sex and gender and research integrity (2020)	1.50
• Journal club (2021-2023)	30.00
• Seminar TUTOR-PHC (2022)	40.00
• Research Integrity Round: Publication ethics. Promises, problems, and perspectives (2022)	1.50
• Research Integrity Round: College tour with Hedi Claahsen, Sandra Heskamp and Bas Bloem (2023)	1.50
Conferences	
• Symposium 'Anders dan normaal' (2019)	3.00
• Symposium - Onderzoek voor iedereen, kwetsbaar maar krachtig (2019)	3.00
• RIHS PhD retreat (2019)	16.00
• Researchdag Associatie van Academische Werkplaatsen VB (2020)	8.00
• Symposium Spoedzorg voor mensen met verstandelijke beperkingen (2020)	5.00
• PhD Day 2020 (2020)	8.00
• PhD Day 2021 (2021)	5.00
• CaRe Days (2021, oral presentation)	10.00
• IASSIDD 6th Europe Conference: Value Diversity (2021, oral presentations)	30.00
• European Public Health Conference (2021, oral presentation)	30.00
• Special Olympics Health Congress (2021)	2.50
• Scientific Seminar (Refereersessie) ELG (2021, oral presentation)	6.00
• Trinity Health & Education International Research Conference (THEconf2022) (2022, oral presentation)	16.00
• RIHS PhD retreat (Apr 2022)	8.00
• RIHS PhD retreat (Nov 2022)	16.00
• NHG wetenschapsdag (2022, oral presentation)	16.00
• Researchdag Academische werkplaatsen VB (2022)	8.00
• WONCA Europe conference (2022, oral and poster presentation)	45.00

Training activities	Hours
• Webinar IASSIDD Health inequities during COVID-19 (2022, oral presentation)	10.00
• Webinar IASSIDD International perspectives on supported decision making (2022)	1.50
• NAPCRG 50th research meeting (2022)	40.00
• CaRe Days (2023)	16.00
• Focus op Onderzoek (2023, oral presentation)	16.00
• NHG Wetenschapsdag (2023, oral presentation)	16.00
• PhD retreat (2023)	16.00
Other	
• Member of RIHS PhD council (2020-2022)	52.00
• Research visit University of Nevada, Las Vegas (2022)	12.00
• Trainee Transdisciplinary Understanding and Training on Research – Primary Health Care (2022-2023)	52.00
• Policy groups and work groups (2020-2023)	36.00

Teaching activities	
Lecturing	
• Werkgroep Onderzoek in VB (2021)	9.00
Supervision of internships / other	
• Co-organising PhD retreats (2021, Apr 2022, Nov 2022)	168.00
• Coaching student internship (12 weeks) (2022)	30.00
• Coaching student internship (4 weeks) (2023)	10.00
• Meet the PhD (2021-2023)	70.00
• Supervising BMW students with grant proposal (2023)	45.00
Total	**1,712.50**

LIST OF PUBLICATIONS
PEER-REVIEWED

van den Bemd, M., Cuypers, M., Bischoff, E. W., Heutmekers, M., Schalk, B., & Leusink, G. L. (2022). Exploring chronic disease prevalence in people with intellectual disabilities in primary care settings: A scoping review. *Journal of Applied Research in Intellectual Disabilities*, *35*(2), 382-398. https://doi.org/10.1111/jar.12957

van den Bemd, M., Schalk, B. W., Bischoff, E. W., Cuypers, M., & Leusink, G. L. (2022). Chronic diseases and comorbidities in adults with and without intellectual disabilities: comparative cross-sectional study in Dutch general practice. *Family Practice*, *39*(6), 1056-1062. https://doi.org/10.1093/fampra/cmac042

Koks-Leensen, M. C., Schalk, B. W., Bakker-van Gijssel, E. J., Timen, A., Nägele, M. E., **van den Bemd, M**., ... & Naaldenberg, J. (2023). Risk for Severe COVID-19 Outcomes among Persons with Intellectual Disabilities, the Netherlands. *Emerging Infectious Diseases*, *29*(1). https://doi.org/10.1093/eurpub/ckab164.835

van den Bemd, M., Cuypers, M., Schalk, B.W.M., Leusink, G.L., Bischoff, E.W.M.A. (2023). Chronic disease management in patients with and without intellectual disabilities: a matched study in Dutch general practice. *British Journal of General Practice, 73*(735), e744-e751. https://doi.org/10.3399/BJGP.2023.0029

van den Bemd, M., Suichies, V., Bischoff, E.W.M.A., Leusink, G.L. (2023). Records of quality indicators for management of long-term health conditions of patients with intellectual disabilities in Dutch residential care. *British Journal of Learning Disabilities*. https://doi.org/10.1111/bld.12570

NOT PEER-REVIEWED

Koks-Leensen, M., Schalk, B., **Van den Bemd, M**., Bakker-van Gijssel, E., Nägele, M., Cuypers, M., Naaldenberg, J., Leusink, G. (2021). Registratie van Covid-19 bij mensen met een verstandelijke beperking. *Tijdschrift voor Artsen Verstandelijk Gehandicapten, 39*(3).

Milou van den Bemd, Bianca W.M. Schalk, Erik W.M.A. Bischoff, Maarten Cuypers, & Geraline L. Leusink. (2022). Chronische ziekten en comorbiditeiten bij mensen met en zonder verstandelijke beperkingen. *Tijdschrift voor Artsen Verstandelijk Gehandicapten, 4,* 155-158.

PUBLISHED CONFERENCE ABSTRACTS

IASSIDD 6th Europe Conference (2021)

Milou van den Bemd, Maarten Cuypers, Erik Bischoff, Marloes Heutmekers, Bianca Schalk, & Geraline Leusink. (2021). Exploring chronic disease prevalence in people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, Special Issue: Proceedings of the 6th IASSIDD Europe Congress: Value Diversity. 34*(5), 1293-1294.

Milou van den Bemd, Maarten Cuypers, Bianca Schalk, Erik Bischoff, Geraline Leusink. (2021). Prevalence of cardiovascular and other chronic diseases in Dutch inhabitants with and without ID. *Journal of Applied Research in Intellectual Disabilities, Special Issue: Proceedings of the 6th IASSIDD Europe Congress: Value Diversity. 34*(5), 1297-1297.

Milou van den Bemd, Maarten Cuypers, Bianca Schalk, Erik Bischoff, Geraline Leusink. (2021). Differences in primary care use and chronic disease management between people with and without intellectual disability es. *Journal of Applied Research in Intellectual Disabilities, Special Issue: Proceedings of the 6th IASSIDD Europe Congress: Value Diversity. 34*(5), 1300-1300.

14th European Public Health Conference (2021)

Koks-Leensen, M., Schalk, B., Bakker-van Gijssel, E., Cuypers, M., **van den Bemd, M**., Nägele, M., ... & Leusink, G. L. (2021). COVID-19 registration in people with intellectual disabilities in Dutch long-term care facilities. *European Journal of Public Health*, *31*(Supplement_3).

van den Bemd, M., Schalk, B., Bischoff, E. W. M. A., Cuypers, M., & Leusink, G. L. (2021). Chronic disease prevalence and patterns of comorbidities in people with intellectual disabilities. *European Journal of Public Health*, *31*(Supplement_3).

27th WONCA Europe Conference (2022)

Milou van den Bemd, Bianca Schalk, Erik Bischoff, Maarten Cuypers, Geraline Leusink. (2022). Chronic diseases and comorbidities in people with vs without intellectual disabilities: Is there need for a different care approach? Available from https://www.woncaeurope.org/file/b4121c25-7b53-4038-a97c-341030afd915/WONCA-Europe-2022-Book-of-Abstracts.pdf, 298-299.

Milou van den Bemd, Maarten Cuypers, Bianca Schalk, Geraline Leusink, Erik Bischoff. (2022). Equal healthcare, worse health: Are vulnerable patient groups seen? Available from https://www.woncaeurope.org/file/b4121c25-7b53-4038-a97c-341030afd915/WONCA-Europe-2022-Book-of-Abstracts.pdf, 711-712.

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