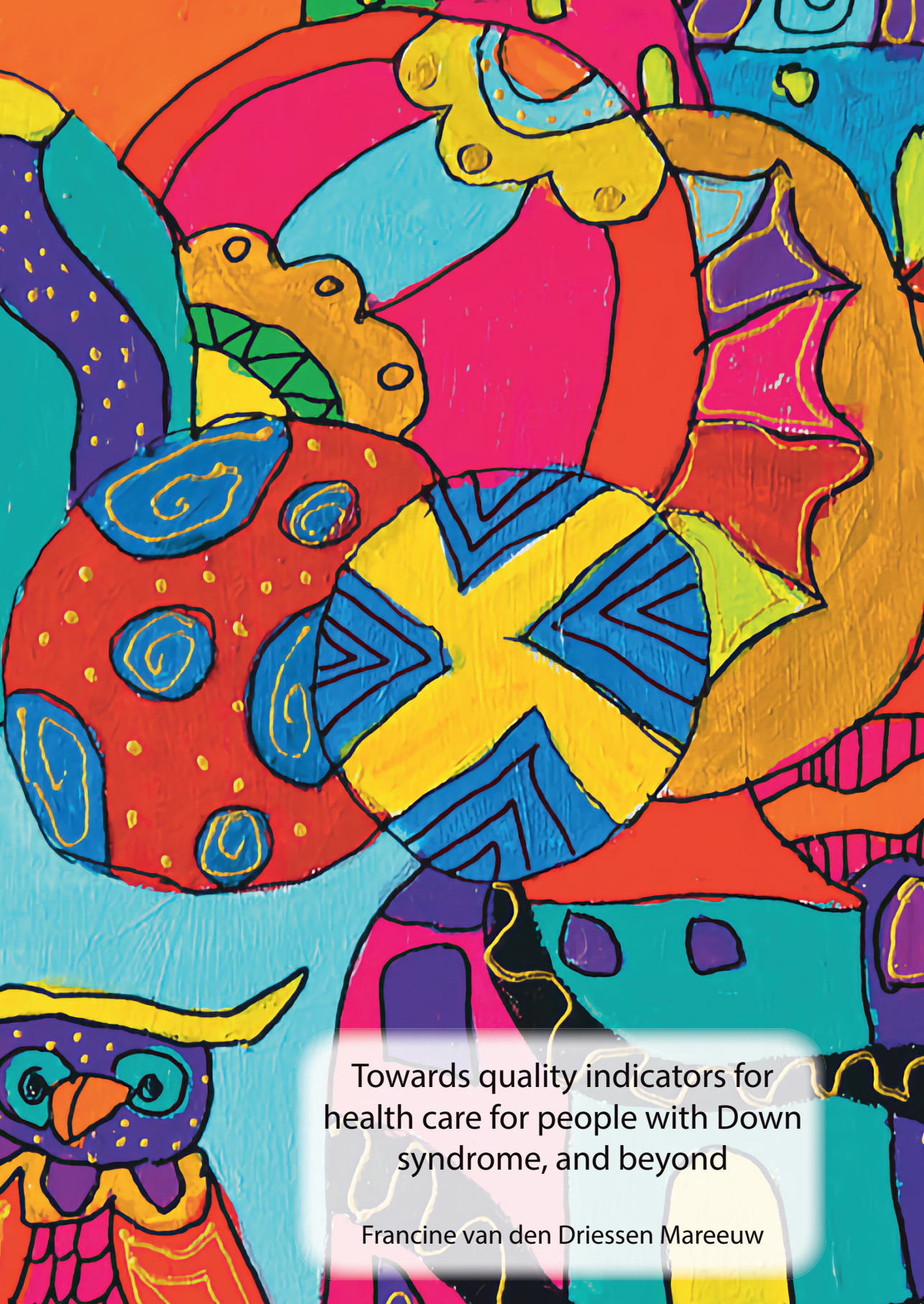

Towards quality indicators for health care for people with Down syndrome, and beyond

Francine van den Driessen Mareeuw

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Proefschrift

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Francine Alexandra van den Driessen Mareeuw,
geboren te Den Haag

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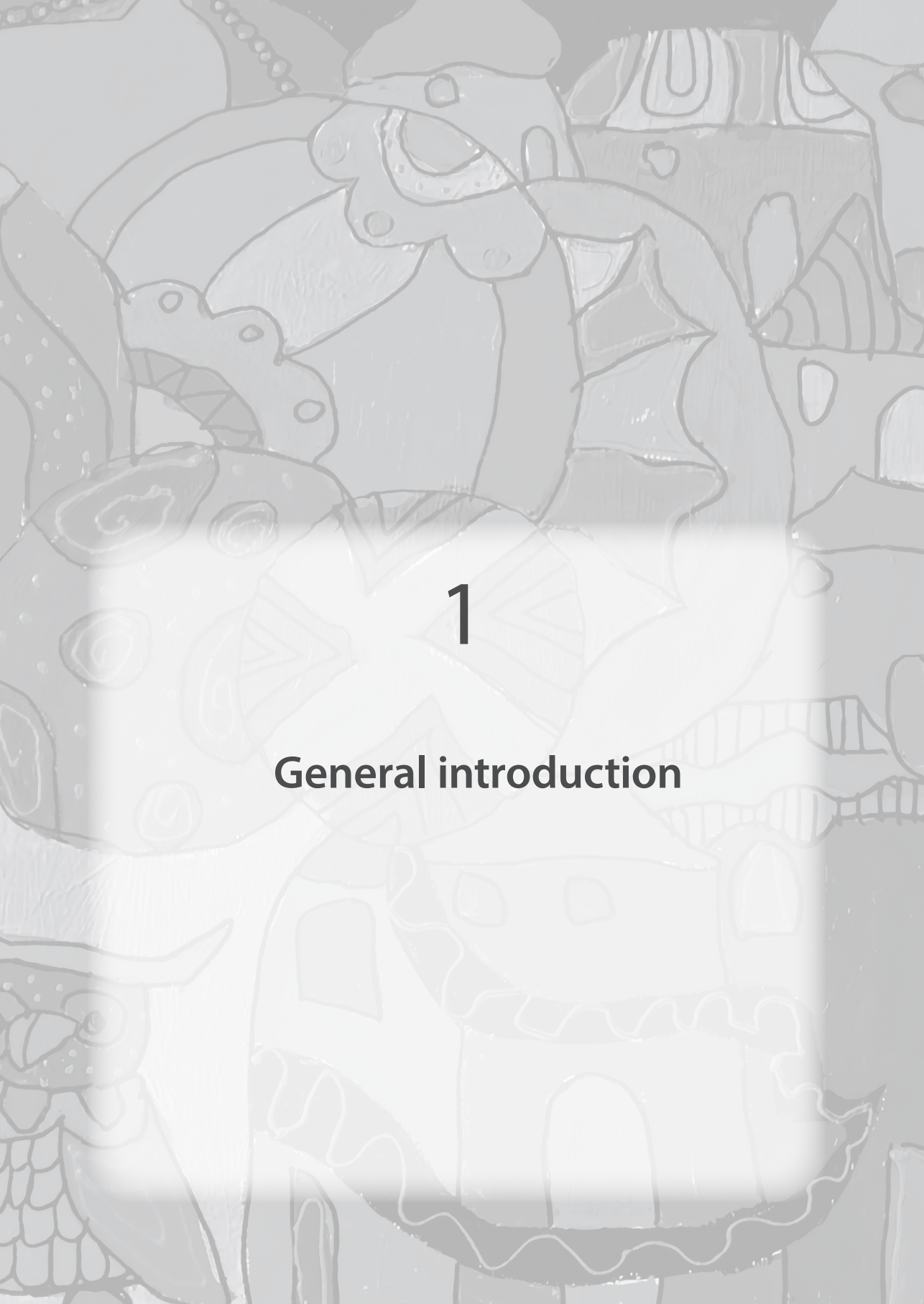

1

General introduction

In short

Down syndrome (DS) is related to intellectual disability and a combination of behavioural patterns and physical health problems (Coppus, 2017; Grieco et al., 2015; Weijerman & De Winter, 2010). As a result of this, people with DS are reliant on a large variety of supports and services. Health care services are one of these. Because of their specific health care needs, high quality health care for people with DS is vital (Kinnear et al., 2018; Skotko et al, 2013). Quality indicators (QIs) can contribute to this quality (Donabedian, 2005). This thesis provides an empirical basis for the content, guidance for the development, and a first draft, of such QIs. A bottom-up approach was applied, which started with the person with DS.

Down syndrome (DS)

Down syndrome was named after J. Langdon Down, who was one of the first, mid 1800s, describing the clinical features of people with the syndrome (Sherman et al., 2007). DS is caused by total or partial trisomy 21 (the presence of a third copy of chromosome 21 or a part of that third copy) (Lagan et al., 2020; Sherman et al., 2007). Maternal age is the major predictive factor for trisomy 21, the chance for having a child with DS increases with maternal age (Sherman et al., 2007).

Number of people with DS

Internationally, the exact number of people with DS is unknown, as reliable registries are scarce (De Graaf et al., 2021; Grevinga et al., 2018; Sherman et al., 2007). Also in the Netherlands, the exact number of people with DS is unknown, as national registries have only started to document data on DS and other congenital anomalies in the 1980s and 1990s (Eurocat, 2021; Grevinga et al., 2018) and these registries may be incomplete and subject to under-registration (Grevinga et al., 2018). However, it is known that life expectancy of people with DS has increased over the past 100 years and is now over 60 years of age (Bittles et al., 2007; Coppus, 2017; De Graaf et al., 2011). An accurate estimate of the population prevalence of DS is essential for an adequate allocation of resources, organisation of care and education, and as grounding for public policy (De Graaf et al., 2021; Sherman et al., 2007). Several attempts were made to estimate the prevalence of DS (Sherman et al., 2007). Using the model by De Graaf et al. (2011), the estimated number of people with DS in the Netherlands was 13.309 in 2015, which corresponds to 7,8 per 10.000 inhabitants (De Graaf et al., 2020). In comparison, the estimated number of people with DS in the whole of Europe was 5 per 10 000 inhabitants

in 2015 (De Graaf et al., 2021) and 6,7 per 10 000 in the US in 2010 (De Graaf et al., 2017). In 2020, the estimated number of people with DS in the Netherlands was 12.690, on a total Dutch population of 17,4 million people (7,3 per 10 000) and is expected to remain stable (De Graaf et al., 2011; De Groot-van Mooren et al., 2021; G. De Graaf, personal communication, October 13 2021). The introduction of non-invasive prenatal testing does not seem to affect the number of live births with DS in the Netherlands (Crombag et al., 2014; De Groot-van der Mooren et al., 2021; Van Gameren-Oosterom et al., 2012). This number of people with DS is substantial. DS is the most common cause of intellectual disability (ID) and people with DS are a relevant subgroup within the group of people with ID (Kinnear et al., 2018; Silverman, 2007; Van Gameren-Oosterom et al., 2013). Furthermore, this relatively large number of twelve to thirteen thousand people with DS in the Netherlands in combination with their specific needs, asks for dedicated means, services and policy (Coppus & Wagemans, 2014; Kinnear et al., 2018).

Conditions related to DS

DS is related to several typical physical conditions, behavioural patterns, and cognitive impairments. However, each person with DS is unique and has his/her own combination of conditions.

Common phenotypic features of DS are a flat nasal bridge, epicanthic folds (skin fold above the upper eye lid covering the inner corner of the eye), and small body length (Bull, 2020; Lagan et al., 2020; Roizen & Patterson, 2003; Weijerman & De Winter, 2010). People with DS have delayed motor development and about half of the people with DS have congenital heart disease, for which they may need surgical correction (Lagan et al., 2020; Van Gameren-Oosterom et al., 2013). Furthermore, hearing and vision disorders, and gastrointestinal and respiratory problems are more common among people with DS as compared to the general population, as well as immune deficits, thyroid malfunction, coeliac disease, leukaemia, skin problems, and overweight (Bull, 2020; De Weger et al., 2018; Lagan et al., 2020; Roizen & Patterson, 2003; Weijerman & De Winter, 2010). Later in life, people with DS may also suffer from premature and accelerated aging (from 40 years of age) and Alzheimer's disease (Bittles et al., 2006; Bull, 2020; Coppus, 2017; Roizen & Patterson, 2003).

Trisomy 21 also causes delayed cognitive development and mild to profound intellectual disability (ID) (de Graaf et al., 2017; Grieco et al., 2015; Patel et al., 2018;

Van Gameren-Oosterom et al., 2013). Delayed language processing and speech problems are also frequent among people with DS (Bull, 2020; Grieco et al., 2015; Patel et al., 2018). Furthermore, people with DS may experience developmental challenges and learning difficulties (Grieco et al., 2015; Roizen & Patterson, 2003). Generally, during adolescence or early adulthood, people with DS reach a level of cognitive functioning, which stabilises and which may gradually diminish later in life (Grieco et al., 2015; Roizen & Patterson, 2003). Nevertheless, people with DS are also known for their strong social skills and ability to copy other people's behaviours (Grieco et al., 2015). Examples of behavioural challenges especially common among people with DS are attention-seeking behaviours, talking to self, noncompliance, wandering, disturbed sleep, and autism spectrum disorders; some of which lessen during adulthood (Bull, 2020; Coppus, 2017; Grieco et al., 2015; Patel et al., 2018; Van Gameren et al., 2013). Depression or compulsive behaviours and dementia are more common among (older) adults with DS (Coppus, 2017).

Needs of people with DS

Given their physical conditions, behavioural and intellectual challenges, people with DS may require a large number and variety of (health) care services and supports in order to live their lives. Regarding the medical domain, they may need general medical care from for instance the general practitioner and dentist, but also speech therapy, physiotherapy, and specialised cardiological care. Additionally, in response to the specific combination of physical conditions, specialised multidisciplinary teams have been set up in many countries (Coppus, 2017; Skotko et al., 2013; Wexler et al., 2009). In the Netherlands, such teams are also present and are called "Downteams": 23 paediatric Downteams and seven adult Downteams or outpatient clinics (Stichting Downsyndroom, 2021). The exact composition of the teams differs, but most of the paediatric teams include a paediatrician, a physiotherapist, a speech therapist, an ear-nose-throat (ENT) physician, audiological screening, and an ophthalmologist. Other disciplines may be consulted based on the needs of the person with DS. A child with DS (with her/his parent(s)) visits all disciplines during one visit. The composition of adult Downteams is also variable, but the adult teams mostly contain an ID physician (a medical doctor specialised in, and trained for, intellectual disability medicine), an ENT-physician, audiological screening, an ophthalmologist, and a dietician. Some adult outpatient clinics are multidisciplinary, but most include an ID physician only, who may consult other professionals if deemed necessary. Paediatric teams

are generally based in hospitals. Adult teams or clinics are based in organisations providing assisted living facilities, and sometimes in hospitals.

Next to these medical services, children with DS may benefit from developmental support, educational support (at home and/or at school) and parenting support (Roizen & Patterson, 2003; Weijerman & de Winter, 2010). In the Netherlands, most children with DS live with their parents and go to regular day care or medical day care centres, and later to regular schools, mostly with extra guidance, or to specialised schools for children with developmental delay. Some children with DS need more intensive care, at home, or in a care home. Adults with DS in the Netherlands either live with their parents, or in an assisted living facility, where they may receive various supports concerning daily living. Furthermore, they go to (sheltered or even paid) work and/or to daily activity centres. Some live and receive (more intensive) care in care homes. Furthermore, people with DS may join sport teams, musical groups, or other activities (mostly for people with ID). The required services and supports of a person with DS in the Netherlands are visualised in Figure 1.1.

According to the Convention on the Rights of Persons with Disabilities (CRPD) (UN, 2006), people with disabilities, including people with DS, have the right to participate in society and live their lives according to their wishes and preferences, and as such, they are entitled to receive all needed care and support to achieve this. The CRPD (article 25) also explicitly addresses the right to the "highest attainable standard of health" (UN, 2006). The Netherlands ratified the Convention in 2016.

Regarding people with DS, the Convention implies that all the above-mentioned services and supports should be in place and of high quality in order to sufficiently answer to the specific needs of people with DS and enable their lives. This is echoed in the literature (Grieco et al., 2015; Kinnaer et al, 2018). Strikingly, although it is clear the special and complex needs of people with DS require tailored care, these needs are not always adequately met (Capone et al., 2018; Phelps et al., 2012).

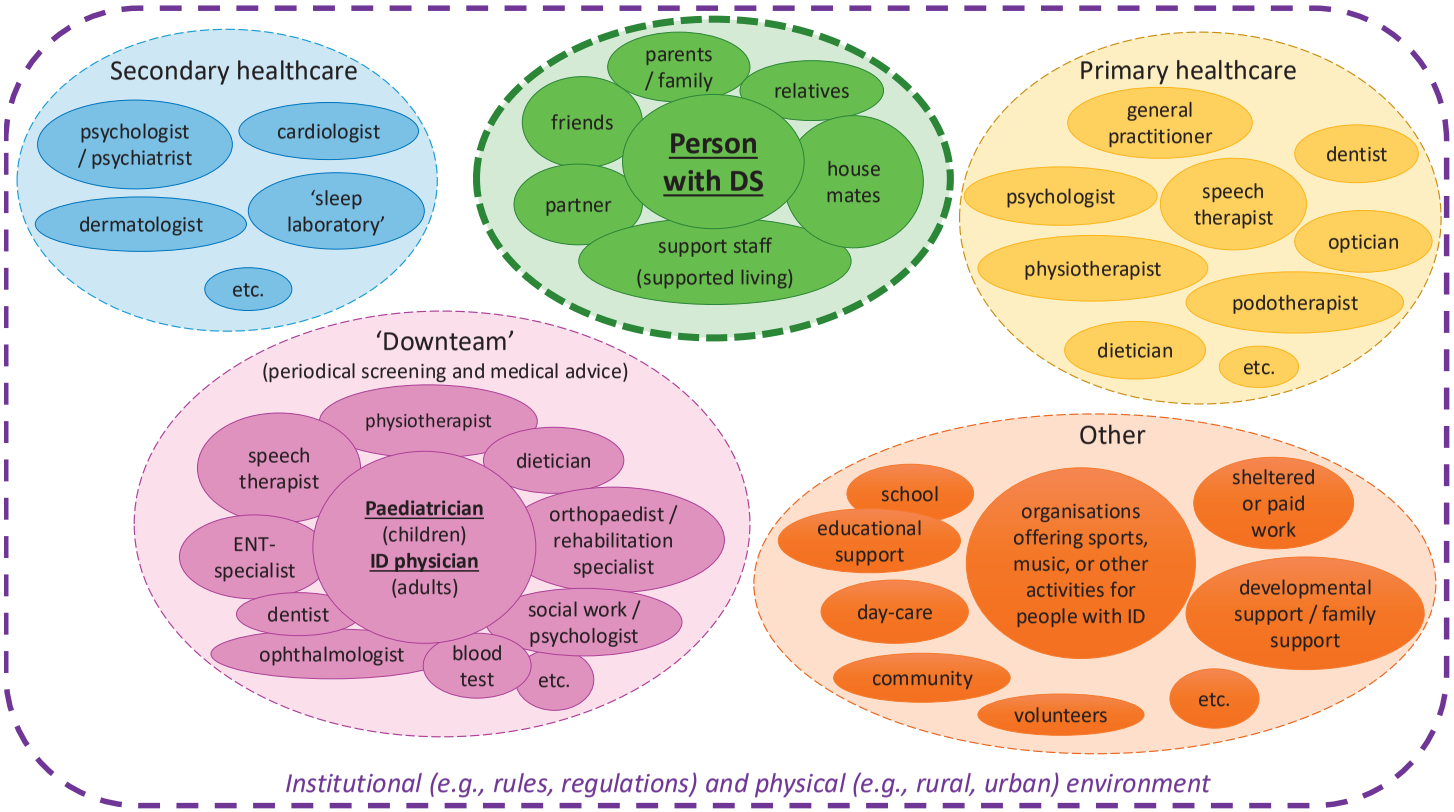

Figure 1.1 Visualisation of potentially required services and supports of a person with DS in the Netherlands

The focus of this thesis: Quality of health care and quality indicators

Although people with DS have this broad spectrum of needs, the focus of this thesis lies on the medical needs and quality of required health care. This focus responds to the identified importance of high-quality health care for people with DS and fills the knowledge gap in this area (Kinnaer et al., 2018; Kyrkou, 2018; Minnes & Steiner, 2009). However, inevitable given the multivariate needs of people with DS, this thesis studies quality of health care for people with DS with the broader picture in mind (taking into account issues beyond the medical domain). By addressing this broader picture, the thesis fits in the current health care landscape, in which increasingly valued principles of person-centred care urge for a more integrated approach (Amalberti et al., 2018; González-Ortiz et al., 2018; Santana et al., 2018). Furthermore, it contributes to the small body of knowledge on health care for people with DS even more, as the little work that has been done on quality of health care for people with DS took the medical perspective only (Jensen & Davis, 2017; Jespersen et al., 2018).

Studying quality of health care starts with the question 'What is quality of health care?'. Many definitions are in use internationally, and they are changing over time (WHO, 2006; WHO, 2018; Busse et al., 2019). Furthermore, the definition may differ depending on where you are located within the health care system and what role you are playing (Donabedian, 1988). In addition, different definitions may be formulated for individuals or populations (Campbell et al., 2000). More concrete, several organisations have formulated quite overlapping dimensions of quality of health care, including (some of) the following (WHO, 2006; WHO 2018; IOM, 2001):

- effective (based on (scientific) knowledge and resulting in best possible health outcomes) (IOM, 2001; WHO, 2006; WHO, 2018),
- safe (e.g., avoiding and minimising injury and risks, utilising safe means) (IOM, 2001; WHO, 2006; WHO, 2018),
- people/person/patient-centred (respectful of, and responsive to, preferences, needs and (cultural) values of the individual and of family and community) (IOM, 2001; WHO, 2006; WHO 2018),
- timely (avoiding waits and (harmful) delays) (IOM, 2001; WHO, 2018),
- equitable (quality of provided care does not differ because of personal characteristics such as age, sex, gender, race, religion etc) (IOM, 2001; WHO, 2006; WHO, 2018),

- integrated (refers to communication between components across the sector, seamless transitions, gaps between clinical settings) (WHO, 2018),
- efficient (maximising resource use and avoiding waste) (IOM, 2001; WHO, 2006; WHO, 2018),
- accessible (timely, geographically reasonable, provided in a setting with appropriate resources) (WHO, 2006).

Generally, in most definitions, components of optimal outcomes for the patient and a firm knowledge base (e.g. evidence-based practice) are prominent (Allen-Duck et al., 2017; Blumenthal, 1996; Campbell, 2000; IOM, 2001; WHO, 2006; WHO, 2018). All of the above-mentioned dimensions are addressed in this thesis.

Also, quality improvements are considered an integral part of quality (Allen-Duck et al., 2017; WHO, 2018). The need for improvements is driven by societal developments such as technical developments, costs, and demographical changes (Amalberti et al., 2019), and is integrated in the daily routine of many health care professionals (Campbell et al, 2003). Quality indicators (QIs), also called quality measures or performance indicators, are important instruments for quality improvement. QIs are measurable and carefully defined items of health care (Campbell et al., 2003; Kötter et al., 2012) and provide insight into health care quality which in turn may identify directions for health care reforms, inform clinical decisions, and help patients finding the needed care (Boulkedid et al., 2011; Campbell et al., 2003; Donabedian, 2005; Rademakers et al., 2011). Generally, three categories of QIs are distinguished: structure, process, and outcome QIs (Donabedian, 1988; Donabedian, 2005). Structure refers to the setting in which health care is provided in terms of material and human resources and organisational structure. Process includes all activities by health care professionals and patients in order to provide and receive care. For example, this involves making diagnoses, but also patient compliance to treatment. Outcome denotes the results or effects of the provided or received care, such as improved health or satisfaction with care.

Despite the growing attention for quality of health care, to date, QIs measuring quality of health care provided to people with DS are scarce (Santoro et al., 2021; Van den Driessen Mareeuw et al., 2017). Up until now, improvement initiatives concerning health care for people with DS are limited to the development of guidelines (Santoro et al., 2016; Tsou et al., 2020; Van Allen et al., 1999). In the

Netherlands, a multidisciplinary guideline for health care for children with DS is present (Borstlap et al., 2011), which is currently revised. A Dutch guideline addressing health care for adults with DS is being developed. QIs providing insight into health care for people with DS are still to be developed. It is the purpose of this thesis to draft such QIs. By doing this, it is the aim to contribute to high quality health care for people with DS, better answer their complex needs and thereby contribute to their quality of life. The latter adds to filling the research gap on the interplay between health care provision and quality of life (Goodman & Brixner, 2013).

Quality of life in itself is another multi-defined and multi-dimensional concept (Eriksson & Lindström, 2007). However, the eight dimensions by Schalock et al. (2005) are considered leading in studying quality of life in people with ID. They include 1) emotional wellbeing, 2) interpersonal relations, 3) material wellbeing, 4) personal development, 5) physical wellbeing, 6) self-determination, 7) social inclusion, and 8) rights.

The QIs drafted in this thesis will be applicable to all primary and secondary health care that people with DS may need during their lives (e.g., health care provided by paediatricians, ID physicians, physiotherapists, dieticians etc. within or outside Downteams). However, the QIs will not address highly specialised, tertiary or academic, care, such as the heart surgery people with DS with congenital heart disease may need. The QIs do cover adequate referrals to such highly specialised care. Furthermore, the thesis seeks to formulate QIs that are relevant to all people with DS, of all ages, and with all combinations of needs.

Research questions

This thesis addresses the following research questions:

1. *What is quality of health care for people with DS?*
   a. *From the 'patient' perspective*
   b. *From the professional perspective*

2. *Which items of quality (quality indicators) provide an adequate indication of this quality?*

3. *What preconditions need to be complied with before the indicators can actually be used?*

4. *To what extent will QIs be able to improve the lives of people with DS?*

This thesis describes the studies that were carried out to answer these research questions. First, we investigated whether QIs for health care for people with DS did already exist. Chapter 2 describes a scoping review searching for existing QIs. After we concluded that such QIs did not seem to exist (we only found one QI measuring thyroid disease in DS in the UK), we started identifying items to be measured by the QIs. A qualitative explorative study identified important elements of health care quality according to people with DS, parents of people with DS, and support staff working in assisted living facilities for people with ID (and DS). This resulted in a first sketch of the QIs and is described in chapter 3. We started with the 'patient perspective' because this perspective is not only considered indispensable for health care improvements (Poitras et al., 2018; Rathert et al., 2012), it is also crucial for responding to patients' needs (Phelps et al., 2012; Trebble et al., 2010). We applied a person-centred approach, in line with current developments in health care, in order to develop QIs that truly matter to people with DS. Not only people with DS ("patients") themselves were included in the study, parents and support staff were also included because they are important members of the social environmental system of a person with DS, which is considered indispensable for obtaining an elaborated view of a person's life (Kyrkou, 2018; Mastebroek et al., 2016; Rawson & Moretz, 2016). Additionally, parents and support staff may function as representatives or interpreters in order to express the opinions of people with DS (Mastebroek et al., 2016). This first sketch based on the patient perspective was then presented to health care professionals working with people with DS and patient organisations during a Delphi-study, which is described in chapter 4. Participants in this study identified desired items for QIs and reflected on prerequisites for future use of QIs. In chapter 5, all collected information was synthesised into concept QIs. Chapter 6 is based on data from the qualitative exploration among people with DS, parents and support staff described in chapter 3, and sets out the broader context of how QIs may, or may not, contribute to people with DS's quality of life. This line of 'putting things into perspective' is continued in the general discussion in chapter 7, which also formulates directions for further steps and implications for practice, policy, and research. Figure 1.2 depicts the steps in the process of drafting the QIs.

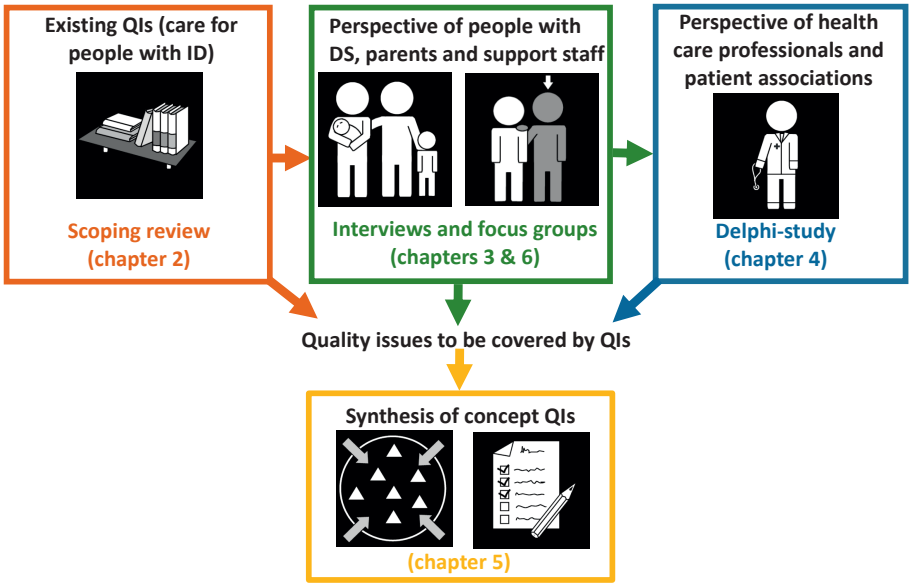

Figure 1.2 Steps of research towards QIs


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In search of quality indicators for Down syndrome health care: a scoping review

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ABSTRACT

Background: The medical care chain around Down syndrome (DS) is complex, with many multidisciplinary challenges. The current quality of care is unknown. Outcome-oriented quality indicators have the potential to improve medical practice and evaluate whether innovations are successful. This is particularly interesting for the evolving care for people with DS and intellectual disabilities (ID). The aim of this study was to identify existing indicators for medical DS care, by reviewing the literature.

Methods: We systematically searched six databases (PubMed, EMBASE, Web of Science, CINAHL, PsycINFO, Google Scholar) for studies concerning the development and implementation of quality indicators for DS and/or ID care, published until February 1st 2015. The scoping review method was used, including systematic data extraction and stakeholder consultation.

Results: We identified thirteen studies concerning quality indicators for ID care that obtained data originating from questionnaires (patient/family/staff), medical files and/or national databases. We did not find any indicator sets specifically for DS care. Consulted stakeholders did not come up with additional indicator sets. Existing indicators for ID care predominantly focus on support services. Indicators in care for people with ID targeting medical care are scarce. Of the 70 indicators within the 13 indicator sets, 10% are structure indicators, 34% process, 32% outcome and 24% mixed. Ten of the thirteen sets include indicators on the WHO quality dimensions 'patient-centredness', 'effectiveness' and 'efficiency' of care. 'Accessibility' is covered by nine sets, 'equitability' by six, and 'safety' by four. Most studies developed indicators in a multidisciplinary manner in a joint effort with all relevant stakeholders; some used focus groups to include people with ID.

Conclusion: To our knowledge, this is the first review that searched for studies on quality indicators in DS care. Hence, the study contributes to existing knowledge on DS care as well as on measuring quality of care. Future research should address the development of a compact set of quality indicators for the DS care chain as a whole. Indicators should preferably be patient-centred and outcome-oriented, including user perspectives, while developed in a multidisciplinary way to achieve successful implementation.

BACKGROUND

Down syndrome (DS), or (partial) trisomy 21, is the most prevalent chromosomal anomaly among new-borns with intellectual disabilities. The overall prevalence throughout the world is about 10 per 10000 new-borns (Roizen & Patterson, 2003; Van Gameren-Oosterom et al., 2012; Weijerman & De Winter, 2010). DS is associated with a broad variety of age-related medical problems, ranging from congenital heart disease to dementia to recurrent respiratory infections (Roizen & Patterson, 2003; Van Gameren-Oosterom et al., 2012; Weijerman & De Winter, 2010). The care chain around a person with DS is challenging and complex, involving numerous professionals (Weijerman & De Winter, 2010; Phelps et al., 2012; Wexler et al., 2009). This requires coordination of care and adequate age- and service-related transitions (Phelps et al., 2012; Wexler et al., 2009).

Initiatives arise to improve the DS care. Skotko et al. (2013) describe how a DS specialty clinic can identify and address many health care needs of children and adolescents with DS beyond the provision of primary care. In the Netherlands, numerous paediatric outpatient clinics now organise such multidisciplinary team appointments, including a visit to the paediatrician, physiotherapist, ENT (ear-nose-throat)-specialist and others, all on the same day. For adults with DS in the Netherlands, health care is less organised, although some 18+ teams are being set up (De Goor, 2011). Internationally, difficulties are identified in care transition (from paediatric to adult care) and in persistent use of paediatric care by DS adults (Jensen & Davis, 2013). An achievement towards higher quality care for DS has been the development of guidelines (Bull, 2011; Borstlap et al., 2011). In general, health checks are increasingly developed in the care for people with intellectual disabilities (ID) (Robertson et al., 2011; Robertson et al., 2014). However, the quality of existing initiatives and the extent to which health care professionals adhere to existing guidelines is unclear (Jensen et al., 2013; Lavigne et al., 2015). More insight is needed into the care that is delivered to people with DS, in terms of types of care, its quality and its effect on clinical outcomes (Lavigne et al., 2015). Quality indicators (also known as quality measures (Boulkedid et al., 2011; Chen et al., 2012)) can provide this insight. They have the potential to structure the development of multidisciplinary teams, improve clinical decisions and guide organisational reform (Donabedian, 2005). This study aimed to review existing data on quality indicators for DS care, including both clinical and organisational aspects, and to identify existing indicator sets.

Evaluating quality of health care (by using indicators) starts with defining 'quality of health care'. About half a century ago (1966) Donabedian formulated the frequently used framework that distinguishes three health care components: *structure, process and outcome* (Donabedian, 2005). Accordingly, the quality of each of these 'care components' can be measured by structure, process or outcome *indicators*. Structure indicators assess the availability of the right facilities, such as staff, supplies, policies and protocols, but also the financial basis, e.g. insurance (Walsh et al., 1999). Process indicators assess whether "good" medical care, according to current evidence/knowledge, has been applied (Donabedian, 2005). Care processes are actions that take place between a patient and care provider, i.e. technical interventions (e.g. measuring blood pressure) or interpersonal interactions (e.g. doctor-patient communication) (Campbell et al., 2000). In practice, process indicators are often operationalised as adherence to guidelines, but they could also include general assumptions like access to and timeliness of services, and coordination and continuation of care. Outcomes are the consequences of delivered care and the actual results of health care interventions, also expressed as the five Ds: death, disease, discomfort, disability and dissatisfaction (Mainz, 2003). Contributions of health care to the patient's quality and length of life may also be qualified as outcomes of health care (Blumenthal, 1996; Campbell & Martin, 2010). Outcome indicators have the potential to evaluate care cycles as a whole instead of single processes by itself (Porter, 2010). Traditionally, measurement instruments (such as indicator sets) for quality of health care contain all three types of indicators (Rademakers et al., 2011).

Next to these three types of health care components, several *quality dimensions* of health care are defined. The World Health Organization (2006) defines six dimensions of quality of care, i.e. care being effective, efficient, accessible, patient-centred, equitable and safe (WHO, 2006). When it comes to *integrated care*, other quality dimensions should be considered as well, such as continuity and adequate transitions between care organisations (Barelds et al., 2010).

Additionally, quality of care can be assessed at different levels, e.g. at the level of single providers, departments, hospitals or at the level of care chains as a whole: the combined efforts of all care providers together (De Koning et al., 2006). In the end, it is this care chain that delivers the total package of care to the patient, resulting in the final outcome (Porter, 2010). Addressing the care chain as a whole

in quality evaluation is quite challenging, because so many organisations and people are involved (Porter, 2010).

In order to contribute to quality improvement, indicators measuring quality of health care should themselves be of good quality, e.g. evidence based, and they should measure what they are designed to measure. An instrument that can be used as a manual to develop indicators is the AIRE instrument (Appraisal of Indicators through Research and Evaluation) (De Koning et al., 2006). In addition, AIRE can be used as a checklist to appraise the quality of indicators (De Bruin-Kooistra et al., 2012).

This study aims to review existing quality indicators for the DS care chain (for both children and adults with DS). We focus on the following research question:

Which indicators are available to assess the clinical and organisational quality of medical DS health care?

More specifically:

1. ***Which indicator sets are available and which indicators do they contain?***
   a. *Which components and levels of care are covered by these indicators?*
   b. *Of which type (structure, process or outcome) are these indicators?*
2. ***What is the quality of these indicator sets?***
   a. *Which dimensions of quality are covered by the sets?*
   b. *How have the sets been developed and implemented?*
   c. *What can be said about other quality aspects of the sets?*

METHODS

A scoping study was carried out to map available indicator sets of health care for people with DS. A scoping study (or scoping review) is a specific type of literature review that may be used to examine research activity in a certain field of study, assess the usefulness of conducting a full systematic review, summarise research findings, or identify gaps in literature (Arksey & O'Malley, 2005; Levac et al., 2010). Scoping studies are often conducted when little research has been done on the topic

studied and a specific research question cannot be formulated (Levac et al., 2010; Victoor et al., 2012). In an attempt to ascertain rigorousness and transparency, Arksey and O'Mally (2005) constructed a framework for conducting scoping studies. The framework consists of five stages: 1) identifying the research question; 2) identifying relevant studies (search strategy); 3) selecting the studies; 4) charting the data (data extraction); 5) collating, summarising and reporting the results; and 6) (optional) consultation of stakeholders, resulting in suggestions for additional references and views (Arksey & O'Malley, 2005; Levac et al., 2010). We followed these stages.

Search strategy

The databases of PubMed, EMBASE, Web of Science, CINAHL, PsycINFO and Google Scholar were systematically searched for articles published until February 1, 2015 (no starting date). These six databases were selected together with a librarian to cover a wide range of biomedical and psychological literature from the perspective of different health care professionals (physicians, psychologists and nurses). The first group of search terms consisted of synonyms for people with DS. The second group of search terms comprised outcomes to target quality indicators, including quality management, quality improvement and benchmarking. Since results for only DS(-synonyms) were very scarce, the first group of search terms was broadened by adding search terms for (synonyms for) people with intellectual disabilities (ID) (Table 2.1). Search strategies were similar for each database, except for Google Scholar, which required a more narrowly defined search, since the entry fields did not accept as many search terms as the entry fields of the other databases.

Table 2.1 Search strategy

Population:	**Outcomes:**
1 Intellectual Disability	11 Quality Indicators, Health Care
2 Mentally Disabled Persons	12 Quality Improvement
3 Developmental Disabilities	13 Total Quality Management
4 Down Syndrome	14 Benchmarking
*5 Developmental disorder**	*15 Clinical indicator**
*6 Mental deficien**	*16 Quality measure**
*7 Mental retard**	*17 Quality assessment**
8 Down's syndrome	
9 Trisomy 21	
10 (1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8 OR 9) (Google Scholar: 1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7)	18 (11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17) (Google Scholar: 11 OR 16)
19 (NOT) Pregnancy	

Combining search term groups: 10 AND 18 NOT 19
This strategy is related to the PubMed search. Very similar versions were used to search EMBASE, Web of Science, CINAHL, PsycINFO and Google Scholar, but adapted for the specific search terms used in these databases, if available. The search terms printed in italics are not MeSH-terms. All MeSH Terms were also searched as free text in all databases as title/abstract.

Study selection

Figure 2.1 shows the selection process in a flowchart. Specific inclusion and exclusion criteria are listed in Table 2.2. In the first selection phase, duplicates were removed, and two independent reviewers (MH or FDM, and EV) screened all titles. Titles were included in the next selection phase when they concerned quality aspects of health care for chronic conditions (comparable to DS care). This review focuses on the care chain for individuals with DS (or ID) from birth to end-of-life. Therefore, we excluded articles concerning prenatal screening. In the next selection phase, abstracts were screened based on more narrow criteria: focus on the development, implementation, application or evaluation of indicators for measuring quality of health care. MH and FDM selected all abstracts (partly by MH, partly by FDM) and a random selection of 30% of all abstracts was screened by a second reviewer (EV, DD, AC, each 10%), which resulted in 26% differences in interpretation. For instance, one abstract mentioned 'Quality deficiencies'; FDM concluded from this that the study was not about indicators, whereas DD thought quality deficiencies could be another word for quality indicators: the study was selected. Another study was not selected, because AC doubted about inclusion (she thought it was not clear whether the study was about health care) and FDM interpreted that the study was not about indicators for health care. Discussion between the reviewers resolved all differences, which resulted in 100% agreement about inclusion or exclusion. MH and FDM reviewed full texts (partly by MH, partly by FDM). In case of any doubt, EV also reviewed the articles and a third and fourth reviewer (DD and AC) was consulted in case of disagreement. In this final phase, quality indicators had to be the main topic, well defined (as well as the population they applied to) and more specifically concerning medical health care, as opposed to e.g. residential care. A snowball method was applied in order to find additional studies: Reference lists of the selected studies were screened for additional relevant studies. If titles mentioned in the reference lists suggested relevant information (on development, implementation or evaluation of indicators), these studies were retrieved and, based on full texts, FDM assessed whether the studies provided additional information. If the studies provided information about additional indicator sets and matched inclusion criteria, these studies were included. If snowball-studies in turn mentioned additional indicator sets in the text, corresponding references were searched too and included if relevant (this happened once).

Table 2.2 Inclusion and exclusion criteria

Inclusion criteria: • Studies concerning the development, implementation, application or evaluation of (structure, process or outcome) indicators for measuring quality of (chronic) medical health care for people with Down syndrome or intellectual disabilities as the main topic • Studies where specific quality indicators are well-defined including the population they apply to • All kinds of scientific publications: journal articles, theses, books, etc.
Exclusion criteria: • Studies where quality indicators itself are not the main topic • Studies not concerning medical care, but other forms of care (e.g. residential care) • Studies concerning general aspects of quality indicators (specific indicators are not well-defined) • Studies concerning quality indicators of general health care (specific population is not described) • Studies primarily focusing on the development of a tool, instrument or questionnaire without the purpose of being an indicator for measuring quality of health care • Studies concerning prenatal or new-born screening/care • No abstract/full text available • Written in a language that no one in the research team masters (i.e. not English, Dutch, French, German)

Data extraction

As the included studies did not always provide enough information to be able to answer our research questions, additional information about the indicator sets was sought. This was done by looking on websites of the organisations who developed the indicator sets and by entering the name of the indicator set in Google and Google scholar.

We extracted data concerning general information about the indicator sets (name of indicator set, author, year, country, target population and organisational context) and about quality domains covered. With the additional information, we were able to assess the indicators in the sets in terms of type (structure, process, or outcome) and quality using the AIRE instrument (mentioned previously). Two researchers (FDM plus EV, DD or AC) appraised each indicator set. The AIRE instrument results in a score for each of its four categories: 1) Aim, relevance and organisational context; 2) Involvement of stakeholders; 3) Scientific evidence; and 4) Further underpinning, formulation and use. For each category, the reviewers need to score several items on a 4-point Likert-scale: 1 meaning not at all agree and 4 meaning very much agree. If no information was available about an item, this was scored as 1. Table 2.3 provides an overview of the four categories of the instrument and of the items per category.

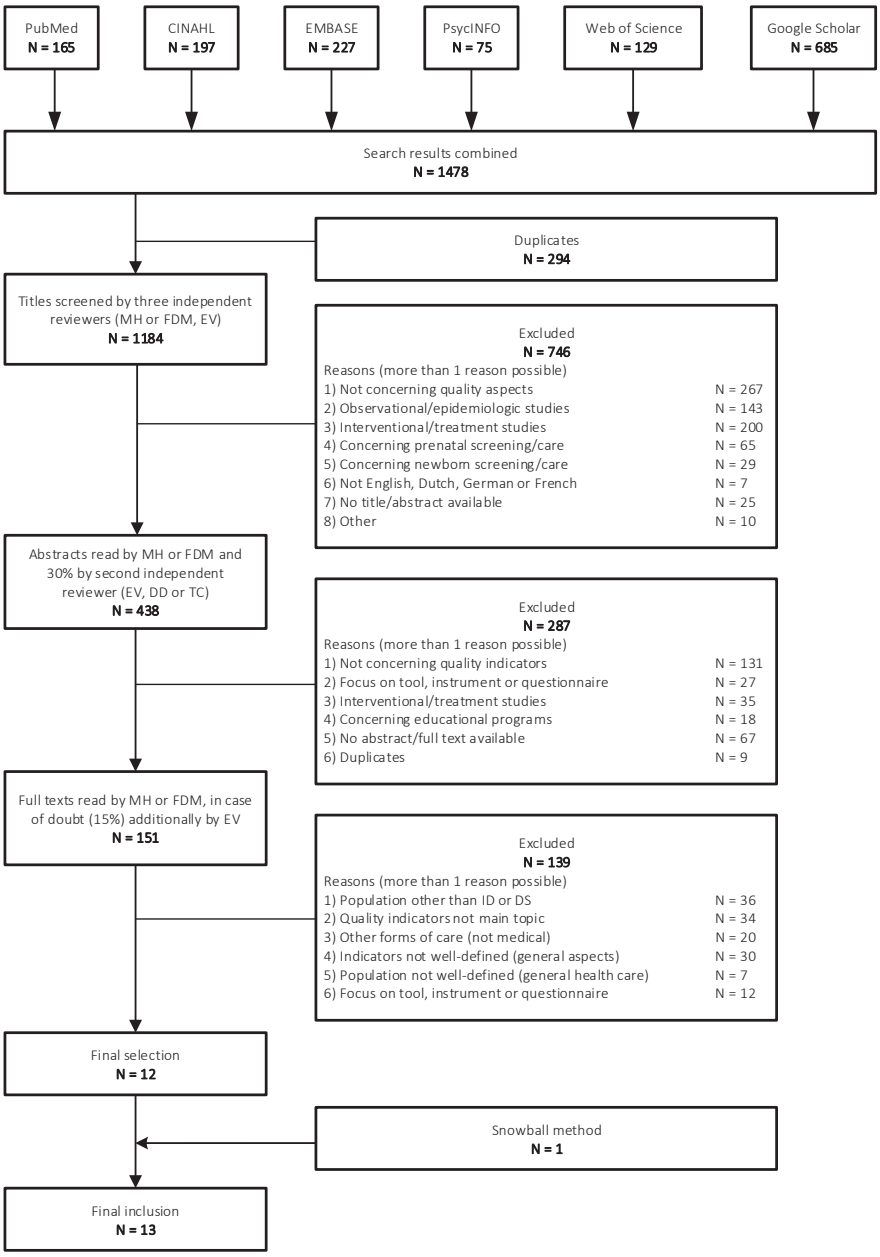

Figure 2.1 Flowchart of selection process
Number of studies found per database, title selection, abstract selection, full text selection, and snowball method resulting in final inclusion of 13 studies.

Table 2.3 AIRE instrument categories and items per category (De Koning et al., 2006)

Categories	Items
1) Aim, relevance and organisational context	- Aim is clearly defined, - Topic relevance is specified, - Organisational configuration (level) is specified, - Quality domain is specified, - Type and size of care process the indicator set applies to is defined.
2) Involvement of stakeholders	- Relevant health care professionals are involved in developing the set, - Relevant other are involved, - The indicator set is formally established (or owned), e.g. by a patient or professional association.
3) Scientific evidence	- Underpinning evidence for the set is systematically searched, - The set is based on a guideline, - The Used evidence is qualitatively good.
4) Further underpinning, formulation and use	- Denominator and numerator are clearly described, - Target population is specifically and clearly defined, - A risk adjustment strategy (for different patient groups) is present, - Validity of the set is proven or argued, - Reliability of the set is proven or argued, - Power of the set is proven or argued, - The set is tested in practice, - The effort needed for data collection is taken into account, - The set includes an instruction for interpretation of the results.

One researcher (FDM) assessed the type of the indicators, as the definition of the types was clear and all indicators could be easily attributed to one of the three types. Some indicators were very broadly defined and were therefore classified as 'mixed', covering information about two or more of the types. For each set, the percentages of the indicator types were calculated, after which the percentages per type were added up in order to provide an idea of relative distribution of indicator types for all the indicators in the sets.

Consultation exercise

Twenty representatives from the health care perspective (professionals providing different sorts of health care to people with DS in the Netherlands) and three from the health care receivers (board members of a leading Down syndrome association in the Netherlands) were asked (by e-mail) to review the list of selected studies and check whether they missed studies or indicator sets. We also asked them about their opinions concerning indicator sets for DS care in general. Four representatives (from the professionals group) did not review the identified studies and indicator sets because of time constraints and/or lack of interest in the topic.

RESULTS

The literature search yielded 1184 studies (see Figure 2.1). No studies specific for DS care were found. Thirteen studies were selected for final inclusion: they contained quality indicators for medical health care in people with ID (see Table 2.5, second column). Consultation of stakeholders did not result in additional studies or indicator sets. All stakeholders agreed that developing indicators for medical care for people with DS would be worthwhile for improving quality or transparency (see Table 2.4).

Table 2.4. Answers of stakeholders

	Number of Times mentioned by stakeholders (n=19)
Why are indicators for DS relevant?	
To define care	8
For coordination	7
For quality improvement	8
For comparability of care providers	14
To check availability	3
Additional studies?	
No	11
Yes but not about indicators	8

Research question 1: Which indicator sets are available and which indicators do they contain?

Thirteen different indicator sets were identified (Table 2.5), five of which originate from the UK, four from the USA, one from Canada, one from Ireland, one from Sweden, and one as a result of a partnership between 13 European countries.

Out of the thirteen identified indicator sets, three have not been specifically developed for people with ID. The three studies describing these sets only evaluated existing indicators in people with ID, by comparison with the general population (no. 9, Quality indicators for preventive care; no. 3, Healthcare Effectiveness Data and Information Set; no. 10, Quality care indicators of diabetes for people with ID). Others adjusted existing sets of indicators to apply them in care for people with ID (no. 1, Ambulatory Care Sensitive Conditions; no. 2, Hospital Admissions for Ambulatory Care Sensitive Conditions; no.5, Measurement of Processes of Care; no. 11, Six Core Outcomes). Three indicator sets have been developed or used for children with, or at risk for, ID, i.e. no. 5 (MPOC-28), no. 9 (Quality indicators for preventive care), and no. 11 (Six core outcomes). An overview of the indicators per set, including their content, can be found as in Appendix I-1.

Table 2.5 Overview of identified indicator sets described by selected studies and general information about the sets

Indicator set	Described by selected study	Country of origin / develop-ment	Target population	Number of indicators (sub-indicators) and Topics covered by indicators in set	Organisational level	WHO quality domains
1 Ambulatory Care Sensitive Conditions (ACSC) (Balogh et al., 2010; Balogh et al., 2011)	Glover & Evison, 2013	Canada	Persons with an intellectual disability	**15:** "conditions which, given 'effective management' at the primary care level, should not normally result in an admission to hospital"	Primary care	Effective, efficient, accessible
2 Hospital Admissions for Ambulatory Care Sensitive Conditions (ACSC) (Glover & Evinson, 2013)	Glover & Evison, 2013	UK	People with learning disabilities (LD)	**3 (22):** Acute conditions, Chronic conditions, immunisable conditions.	National health system of England	Effective, efficient, accessible
3 Healthcare Effectiveness Data and Information Set (HEDIS®) (Shireman et al., 2010; NCQA, 2015a; NCQA, 2015b)	Shireman et al., 2010	USA	Adults with developmental disabilities with Diabetes	**5:** HbA1c testing, eye examinations, lipid testing, microalbuminaria screening, primary care visits	National/ whole care chain	Effective, patient-centred
4 The Health Equalities Framework (HEF) (Thomas, 2014; Atkinson et al., 2013)	Thomas, 2014	UK	People with learning disabilities (LD)	**5 (29):** Social indicators, Genetic and biological indicators, Communication difficulties and reduced health literacy indicators, Personal behaviour and lifestyle indicators, Deficiencies in service quality and access indicators	Specialist multidisciplinary learning disability services	Efficient, accessible, patient-centred, equitable, safe
5 Measurement of Processes of Care (MPOC-28) (Granat et al., 2002; Cunningham & Rosenbaum; 2014)	Granat et al., 2002	Sweden	Families with children with disabilities	**4 (28):** Enabling and partnership, General & specific information (given by care provider), Co-ordinated and comprehensive care, Respectful and supportive care	Child rehabilitation services departments	Efficient, accessible, patient-centred

Table 2.5 continued

Indicator set	Described by selected study	Country of origin / develop-ment	Target population	Number of indicators (sub-indicators) and Topics covered by indicators in set	Organisational level	WHO quality domains
6 National Core Indicators (NCI) (Bradley et al., 2007; NCI, 2015a; NCI, 2015b)	Bradley et al., 2007	USA	Children and adults with developmental disabilities and their families	**5 (94):** Individual outcomes (satisfaction, choice and decision making, self-determination, community inclusion, work, relationships), Health welfare and rights (safety, health, medication, wellness, restraints, respect/rights), System performance (Service coordination, Access, staff stability), Family indicators (choice & control, family outcomes, information & planning, satisfaction, family involvement, community connections, access & support delivery).	Public systems for people with intellectual and developmental disabilities	Accessible, patient-centred, equitable, safe
7 Quality Indicators ~ February 2004 Learning Disabilities (NHS-QIS) (NHS Quality Improvement Scotland, 2004; N HS Quality Improvement Scotland, 2006)	Campbell, 2008 NHS Quality Improvement Scotland, 2004	UK, Scotland	Children and adults with learning disabilities in Scotland	**6 (60):** Involvement of Children and Adults with Learning Disabilities and Their Family Carers through Self-Representation and Independent Advocacy, Promoting Inclusion and Wellbeing, Meeting General Healthcare Needs, Meeting Complex Healthcare Needs, In-patient Services - Daily Life, Planning Services and Partnership Working	National Health System of Scotland	Effective, efficient, accessible, patient-centred, equitable, safe
8 Health indicators for people with intellectual disabilities (Pomona-project) (POMONA II Research group, 2006; Van Schrojenstein Lantman-de Valk, 2007)	van Schrojenstein Lantman-de Valk et al, 2007 (snowball)	Europe	People with intellectual disabilities in Europe	**4 (18):** Demographics, Health status, Determinants of health, Health systems.	European/national	Effective, efficient, patient-centred, equitable

Table 2.5 continued

Indicator set	Described by selected study	Country of origin / development	Target population	Number of indicators (sub-indicators) and Topics covered by indicators in set	Organisational level	WHO quality domains
9 Quality indicators for preventive care (Coker et al., 2012; Blumberg et al., 2012; NSCH, 2007)	Coker et al, 2012	USA	Children aged 10 months to 5 years old who are at risk for developmental delay	**4 (14):** Parents' Evaluation of Developmental Status, Comprehensive and coordinated care, Family-centred and culturally effective care, medical home.	Preventive care	Effective, efficient, accessible, patient-centred
10 Quality care indicators of diabetes for people with ID (Diabetes UK, 2014; Taggart et al., 2013)	Taggart et al, 2013	UK	People with intellectual disabilities and diabetes	**1(6):** HbA1c checked, Lipids/cholesterol, Eye exam, Weight change, Physically active, Attended emergency department related to DM	Diabetes care chain	Effective, efficient, patient-centred
11 Six Core Outcomes: Key Measures of Performance (Blumberg et al., 2008; NS-CSHCN, 2009; Spears, 2010; Strickland et al., 2011; US Department of health and human services, 2007)	Spears, 2010	USA	Children with special health care needs	**6:** Shared decision making, Coordinated care, Adequate insurance, Screening for special health care needs, Community-based services, Services for transitions.	States' and Territories' service systems	Effective, efficient, accessible, patient-centred
12 Quality and Outcomes Framework Indicators for learning disabilities (QOF) (Ashworth, 2012; NICE, 2009; NICE, 2010; NICE, 2013; NICE, 2016a,b,c)	Ashworth, 2012	UK	People with learning disabilities in the UK	**1(2):** Learning Disability register, % Patients in register with Down's Syndrome aged 18 and over who have a record of blood TSH in the previous 15 months.	Primary care	Effective, efficient, equitable
13 Quality indicators measuring the quality of the medication use process for people with intellectual disabilities (Flood & Henman, 2015; Flood & Henman, 2016)	Flood & Henman, 2016	Ireland	People ageing with intellectual disabilities	**5 (37):** Patient experience, access to care, continuity of care, equity, patient safety, effectiveness, appropriateness, assessment.	Medication use process care chain	Effective, accessible, patient-centred, equitable, safe

Research question 1a: Which components and levels of care are covered by the indicators?

The indicator sets cover a large variety of health care levels (settings) and topics. The sets predominantly evaluate the presence of facilities/services or the effectuation of care delivery at communicational and organisational levels. Most of the sets include indicators on collaboration, multidisciplinary cooperation, transition and coordination. Five of the identified sets focus on quality of supportive care and services, containing only a subcategory of indicators being applicable to medical care: no. 3 (The Health Equalities Framework, HEF), no. 6 (National Core Indicators, NCI), no. 7 (the NHS quality indicators for Learning Disabilities, NHS-QIS), no. 9 (the Quality indicators for preventive care), and no. 11 (the Six Core Outcomes). Medical care is approached in a general way and specific diseases and/or treatment courses are barely addressed. Indicators on medical topics primarily focus on screening and preventive care. Two sets consider hospitalisation rates as indicators for conditions which, given effective primary care, should not normally result in hospital admission. Their indicators aim to measure access to, and quality of, primary care: no. 1 (Ambulatory Care Sensitive Conditions) and no. 2 (Hospital Admissions for Ambulatory Care Sensitive Conditions). One set, no. 12 (Quality Outcomes Framework, QOF) contains - among others - an indicator named 'Learning disabilities', which comprises a measure for a register of patients with learning disabilities and a measure for thyroid disease among people with DS (NICE, 2015). This is the only set explicitly addressing DS. The QOF indicators have been designed to measure the quality of primary care in Great Britain. Two indicator sets include measures for diabetes care for people with intellectual disabilities (no. 3, Healthcare Effectiveness Data and Information Set; no. 10, Quality care indicators of diabetes for people with ID). Lastly, two sets focus on processes of care: i.e. no. 5 (MPOC-28) concerning processes in child rehabilitation and no. 13 (Quality indicators for medication use process) including indicators for medication use in people with ID.

Research question 1b: Of which type (structure, process and outcome) are the indicators?

The number of indicators per set varies widely. The thirteen sets together comprise 70 separate indicators, ranging from 2 to 6 indicators per set. Most indicators in turn consist of a number of sub-indicators ranging from 14 to 94. Altogether (regardless of sub-indicators) we identified 6 structure, 21 process, 26

outcome indicators, and 12 indicators measuring a mix of structure-, process-, or outcome-measures. When calculating the percentages of types of indicators per sets, and then adding up the percentages per type, it appeared that 10% of the 70 indicators are structure indicators, 34% process, 32% outcome and 24% mixed. Table 2.6 presents the distribution of the types of indicators per set.

Table 2.6 Relative and absolute proportion of types of indicators in identified indicator sets

type of indicator→ Indicator sets ↓	Structure	Process	Outcome	mix
1 ACSC Can	0	0	100% (15)	0
2 ACSC UK	0	0	100% (3)	0
3 HEDIS DM	0	100% (5)	0	0
4 HEF	0	40% (2)	20% (1)	40% (2)[a]
5 MPOC-28	0	100% (4)	0	0
6 NCI	20% (1)	20% (1)	20% (1)	40% (2)[b]
7 NHS-QIS	33% (2)	17% (1)	0	50% (3)[c]
8 Pomona	0	0	75% (3)	25% (1)[d]
9 Preventive care	0	75% (3)	25% (1)	0
10 Diabetes UK	0	0	0	100% (1)[e]
11 Six Core Outcomes	33% (2)	67% (4)	0	0
12 QOF	50% (1)	0	50% (1)	0
13 Medication use process	0	20% (1)	20% (1)	60% (3)[f]
Total	86 (6)	439 (21)	420 (26)	315 (12)

[a] Mixed indicators consisted of a mix of 1) structure & outcome sub-indicators and 2) structure & process sub-indicators.
[b] Mixed indicators consisted of a mix of 1) structure & process & outcome sub-indicators and 2), structure & process sub-indicators.
[c] Mixed indicator consisted of a mix of structure & process sub-indicators.
[d] Mixed indicator consisted of a mix of structure & process sub-indicators.
[e] Mixed indicator consisted of a mix of process & outcome sub-indicators.
[f] Mixed indicators consisted of a mix of 1) process & outcome sub-indicators (2x) and 2), process & outcome & structure sub-indicators.

Research question 2: What is the quality of the indicator sets?

The quality of the indicator sets was assessed using the AIRE instrument. The AIRE-scores are presented in Figure 2.2.

Although category 1 did not get the highest score in all sets (sets 1, 7, 8, 9, and 11 got a higher score on category 2 and set 5 on category 3), category 1 is the best scoring category on average. All sets have clearly defined the aim and relevance and specify the organisational configuration, type of care, quality dimension on which the indicators apply, and indicate the relevance of the topic. All WHO quality dimensions (effective, efficient, accessible, patient-centred, equitable and safe) are covered (Table 2.7), although some dimensions are only covered by a small

number of sets (e.g. only four indicator sets cover 'safety'). The domains 'effective', 'efficient', and 'patient-centred' are covered by ten of the sets. This implies that a large part of the indicator sets aim to measure (and improve) these dimensions of care. 'Accessibility' is covered by nine sets, 'equitability' by six, and 'safety' by four.

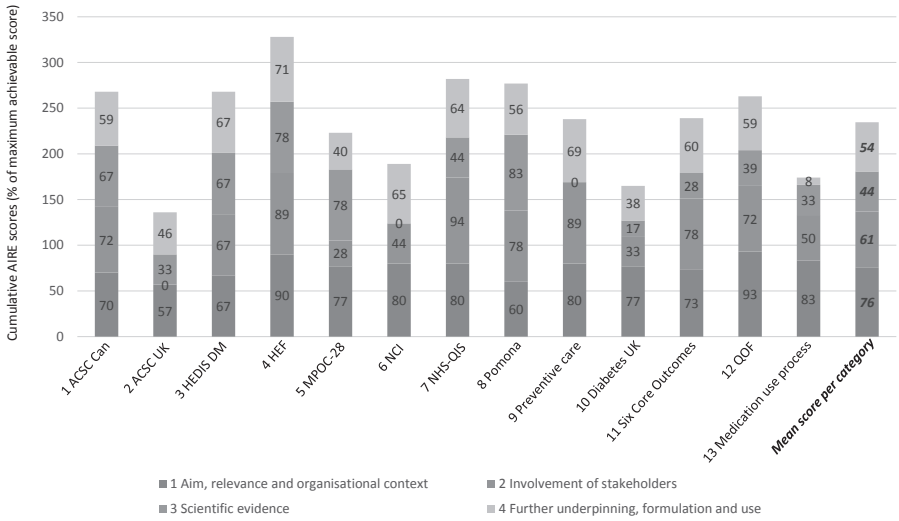

Figure 2.2 AIRE-scores per set.

Scores are calculated as percentage of maximal achievable score. Each colour in a bar reflects the score for an AIRE-score category.

Table 2.7 Quality dimensions covered by indicator sets, per dimension

Quality dimension → Indicator sets ↓	Effective	Efficient	Accessible	Patient-centred	Equitable	Safe
1 ACSC Can	√	√	√			
2 ACSC UK	√	√	√			
3 HEDIS DM	√			√		
4 HEF		√	√	√	√	√
5 MPOC-28		√	√	√		
6 NCI			√	√	√	√
7 NHS-QIS	√	√	√	√	√	√
8 Pomona	√	√		√	√	
9 Preventive care	√	√	√	√		
10 Diabetes UK	√	√		√		
11 Six Core Outcomes	√	√	√	√		
12 QOF	√	√			√	
13 Medication use process	√		√	√	√	√
Number of sets covering dimension	10	10	9	10	6	4

In general, there are differences in whether relevant stakeholders have been involved in developing the sets (AIRE-category 2). In most studies, indicators have been developed in a multidisciplinary manner with involvement of the relevant stakeholders. These stakeholders involve general practitioners, paediatricians, psychologists, social workers, direct care staff, researchers, policy makers, managers and/or family members. In most cases, the actual content of the multidisciplinary team is not clearly described. Two studies have been using focus groups to include people with ID in the development process (Atkinson et al. 2013, and Van Schrojenstein Lantman-de Valk et al. 2007). Other ways of obtaining data for the development of indicators include Delphi studies, web-based applications, on-site observations, staff questionnaires, medical file recordings, financial registrations, content of protocols and/or national databases.

The evidence base of the sets, category 3, provided the lowest scores, though some sets score quite high (no. 1, 3, 4, 5 and 8).

Finally, category 4 (Further underpinning, formulation and use) covers a large variety of indicator characteristics (see Table 2.3) and the score for this category differs between the sets. Some of the sets do not contain indicators with a numerator and denominator, e.g. the two sets on diabetes care contain the indicator 'patient's HbA1c is checked'. Furthermore, some sets clearly report how validity and reliability have been assured, while others do not contain any information on that. The same is true for the power of the sets (the extent to which an indicator is sensible to measure changes). Almost all sets have to some extent been implemented and tested in practice. However, some sets have only been implemented and tested once, while others have been in use for many years. Data collection of the indicator sets also varied. For three of the sets, data collection methods are not (yet) specified (sets 1, 4 and 13). Six of the sets (sets 5, 6, 8, 9, 10, and 11) collect data through telephone surveys, postal questionnaires or face-to-face interviews with people with ID or their representatives. Three sets use existing registrations for obtaining data (2, 3, and 7). For one set (12), general practices have to score points on several topics, it is unclear whether this is done through a questionnaire or existing registrations.

DISCUSSION

Summary of results

We reviewed the literature to identify indicators that assess the clinical and organisational quality of medical care for people with DS. No studies specific for DS care were found (although one study described an indicator set containing one single indicator on thyroid disease among people with DS). Therefore, we have chosen to search for quality indicators in care for people with ID that could be applicable in DS care. We have found that quality indicators in care for people with ID targeting medical care, instead of supportive care and services, were scarce. We reviewed to what extent these indicators cover the structure, process and outcome of care. The majority of indicators concern processes of care for performance measurement. Many sets include indicators on coordination, multidisciplinary working and cooperation. The six WHO quality dimensions are well covered by the sets, although 'safety' is the least addressed. We also aimed to evaluate the development and implementation of the indicators. Most quality indicators have been developed in a multidisciplinary manner with relevant stakeholders, some using focus groups to include people with ID. Almost all sets have to some extent been implemented and tested in practice. Data collection for the indicators is achieved in multiple ways, such as consumer/family surveys, medical file recordings, and/or national databases. The sets differ in quality aspects, e.g. some authors describe thoroughly how validity and reliability was assured, how sensible the indicators are and what the evidence base is, while others barely address these issues.

Quality indicators in medical care for people with ID and DS

The most striking finding of the current study is that quality indicators specific for DS care have not been published to date (except for the single set containing one indicator on thyroid disease among people with DS). Moreover, the indicators found for the care for people with ID barely address medical aspects. Generally, people with DS and people with ID have similar health needs (Phelps et al., 2012), which may imply that the identified quality indicators would be applicable in DS care as well. However, people with DS usually have more and many specific comorbidities compared to the general population of people with ID (Phelps et al., 2012). This urges the need for both medical care that is specifically tailored to the health care needs of people with DS and DS specific indicators, which can

contribute to the quality of life of people with DS (Skotko et al., 2013). Indicators for care for people with ID would not be specific enough. DS specific indicators can reveal bottlenecks in the care chain and can lead to the identification of successful interventions and contributors to a specific outcome (Porter, 2010).

The high prevalence of comorbidities among people with DS also requires multidisciplinary collaboration and coordination. Many of the indicator sets found in this study contain indicators for these requirements. They are general concepts that are applicable to different health care sectors, regardless of the patient group. Thus, regarding multidisciplinary collaboration and coordination, the identified indicators could be used in a set for health care for people with DS.

The six WHO quality dimensions could also be used to define potential indicators (WHO, 2006). In this study we found that the dimensions 'effective', 'efficient', and 'patient-centred' are predominantly covered (ten out of thirteen), while improvement of care – addressing total care chains – should always be done by paying attention to all the six dimensions (WHO, 2006). Nonetheless, we believe that 'equitability' and 'patient-centredness' should receive special attention in DS. People with DS experience inequality in received health care (Henderson et al., 2007). The comorbidities, communication difficulties caused by intellectual disability, and unusual presentation of common diseases of people with DS require more effort from health care professionals to deliver good care (Skotko et al., 2013).

Structure, process or outcome of care

Of the indicator sets we found in this study, many consist of a large number of process indicators. Outcome indicators also comprise a significant part (although less than process) of the indicators in the sets. The number of structure indicators is the lowest. The different types of indicators may be used for different reasons.

Many organisations focus on the assessment of structural aspects and service delivery for performance measurement. They seem to assess results that are easy to reach and easy to measure, with data readily collectable (Campbell et al., 2000; Porter, 2010; Alonazi & Thomas, 2014; Van Loon et al., 2013). Structural aspects of care are essential, as they are the basis of the health care system. Structure indicators are based on the assumption that given the presence of right physical

or staff characteristics, good care automatically results (Donabedian, 2005). However, focusing merely on the structural context as an end in itself, may result in overshadowing the initial goal of improving health outcomes for patients (Alonazi & Thomas, 2014).

Process indicators are based on *how* health care is delivered, e.g. coordination, timeliness, interactions, and *what* interventions take place, e.g. screening or diagnostic tests, treatment etc. Measuring processes has several benefits: they can be measured on a short-term (i.e. directly after care has been delivered), data are easily obtained and differences between organisations are relatively easy to interpret. In general, process indicators are largely based on (the adherence to) guidelines, consisting of recommendations based on current evidence, or best knowledge. Measuring the adherence to guidelines results in important information on the feasibility of recommended care and to some extent, information on care quality. However, standards of best clinical practice are not stable and almost never final (Donabedian, 2005). When we solely measure processes we might risk anchoring what is currently known as best practice, which might result in ceasing of innovation (Porter, 2010).

Outcome indicators measure the consequences of delivered care and actual results of health care interventions. They reflect whether structural context and processes in single organisations, as well as total care chains (Mainz, 2003), actually lead to health benefits. This information on desired, as well as detrimental outcomes may stimulate innovation through the identification of its contributing factors (Porter, 2010). Outcomes can therefore be interpreted as fundamental measures for quality of health care.

Developing an indicator set for DS

According to the above, development of indicators for medical care should focus on developing outcome indicators. There are however some considerations that should be taken into account. Firstly, stakeholders may have different views on which outcomes are desirable. Whereas survival may be the best scenario in the eyes of a physician, a patient may choose functional status above life expectancy. In addition, change in health-status may not always be the primary goal, especially in long-term care (Barelds et al., 2010), support and processes of care may be of greater importance. Indeed, when evaluating user perspectives on this topic, users

primarily seem to focus on processes of care or procedural outputs (Rademakers et al., 2011; Barelds et al., 2010). As patients are the experts when it comes to their outcomes, it is essential to include people with DS and/or their parents in the process to define what is valuable to them (Wiering et al., 2016). Their views on quality differ from those of professionals and researchers (Barelds et al., 2010). Physicians and all other professionals, including health care managers, should also be involved, since they might appraise the usefulness and quality of indicators in a different manner (Campbell et al., 1999). By involving all stakeholders in the development process, their conflicting interests can be identified and weighed against each other. We also saw this stakeholder involvement in the development of many of the identified indicator sets. Defining potential quality indicators for DS should thus involve all relevant stakeholders (De Koning et al., 2006; Flood et al., 2014) (e.g. general practitioners, paediatricians, psychologists, social workers, direct care staff, researchers, policy makers, managers and family members).

Secondly, another consideration when developing outcome indicators is that before outcomes become manifest, long periods of time may elapse and data will not be readily available (Donabedian, 2005; Campbell et al., 2000; Porter, 2010). Therefore, long-term measures should be accompanied with intermediate, short-term outcomes (Mainz, 2003).

Thirdly, as stated before, multidisciplinary working is of vital importance in medical care for people with DS. Moreover, Callaghan (2006) argues that, especially for people with ID, multidisciplinary collaboration leads to better personal outcomes. This would be a reason for including process indicators, since multidisciplinary working is a typical process aspect of care. On the other hand, as multidisciplinary working leads to personal outcomes, outcome indicators may also be suitable to measure quality of care. In any case, multidisciplinary collaboration should be taken into consideration, whether it is measured by process or outcome indicators.

Fourthly, patient characteristics and environmental factors, e.g. intrinsic motivation or socio-economic status, have an important role in influencing health outcomes as well, beyond the control of individual health professionals (Campbell et al., 2000), not to mention comorbidity. Hence, adjusting for this kind of factors outside the health care system that may influence health outcome is important when it comes to interpreting outcomes data (Mainz, 2003). It has to be identified

what exactly leads to the result that is measured. Clinical expertise is needed for adequate interpretation, though what the expected outcomes are, is not always known (Donabedian, 2005).

Finally, when developing indicators one should consider that health care systems differ per country or state (Campbell et al., 2000). Indicators should fit in the care system they apply to. In the Netherlands for example, some DS specific initiatives have been developed. However, specialised care for adults with DS is still scarce (De Goor, 2011). Structural indicators may help in the development of this care, by defining what structural components of care are needed.

To conclude, quality indicators for medical DS care should focus on outcomes, with the above considerations advocating the additional use of some process and structure indicators.

Strengths and limitations

To our knowledge, this is the first review that searched for studies on quality indicators in DS care. With the use of six different databases, we covered a wide range of scientific publications. Moreover, this review discusses strategies for future development of indicators. The study contributes to existing knowledge on DS care as well as on measuring quality of care for other chronic conditions. A strength of the study is the consultation of relevant stakeholders as a last step of the review, which enabled us to check whether we had missed relevant studies or indicator sets. The fact that no additional indicator sets or studies came up in the stakeholder consultation, shows that we did not miss studies and advocates the quality of this review. Additionally, all stakeholders considered development of quality indicators for care for people with DS relevant, which also indicates the relevance of this study.

This study yielded no indicator sets on medical DS health care and the found indicator sets for ID health care predominantly focus on non-medical care (e.g. supportive care). This may be the result of including (synonyms for) intellectual disabilities as a search term, which may have put an emphasis on cognitive disability, which is not necessarily related to medical care. Using search terms on for example congenital abnormality or genetic defects might have possibly yielded more medical studies. However, these studies might have been too

general and less applicable to DS. As ID is one of the outcomes of DS, we chose to search for studies on ID.

A limitation of the study was that the information of the identified indicator sets was somewhat incomplete. We only searched for information through the internet. Due to this incomplete information, not all items of the AIRE instrument, used to assess the quality, could be scored by the reviewers. Therefore, the low AIRE scores, especially regarding the evidence base of the sets, do not necessarily mean that the evidence base of the sets is not good. The low scores may also be a result of little available information on the sets. Consulting organisations that had developed the indicator sets might have yielded more information. However, the number of items with missing information is small and without the AIRE-scores, we are still able to show information on quality (development, implementation, quality domains).

Conclusions

This review gives an overview of different strategies for quality measurement. Quality indicators specific for DS care have not been published to date and in the found studies about the care for people with ID medical aspects are barely addressed. Quality indicators can play a major role in improving medical practice and evaluating whether innovations are successful. This is particularly interesting for the evolving DS care, as well as care for people with ID. As illustrated in this review, it is very hard to focus on specific care quality aspects, when approaching such a diverse, large group as 'people with intellectual disabilities'. Therefore, we recommend focussing on well-defined, DS-specific care chains when developing indicators. Further research activities should include the preparation and development of a compact set of indicators to evaluate and monitor the quality of the DS care chain as a whole. Future indicators should preferably be patient-centred and outcome-oriented, including user perspectives. In order to achieve successful implementation, it is crucial that all care providers support the indicator set, and that all care providers, patients (and/or their parents), and health care managers are involved in the process of development.

APPENDIX

Appendix 1-I

Separate indicators per set and topics covered by indicators.

Indicator numbers correspond to the indicator set they belong to. For example: 2.1 means: indicator set number 2, first indicator; 1.0 means: indicator set number one, only indicator in set.

No.	Indicators and sub-indicators	type
1.0	Ambulatory Care Sensitive Conditions: Asthma; Angina Pectoris; Congestive heart failure; Gastrointestinal ulcer; Immunization preventable infection; Malignant hypertension; Otitis Media; Neurotic depressive disorders; Dental conditions; Diabetes Mellitus; Pelvic inflammatory disease; Constipation; Gastroesophageal reflux; Epilepsy; Schizophrenic disorders.	Outcome
2.1	Acute conditions: Cellulitis; Convulsions and epilepsy; Dehydratation and gastroenteritis; Dental conditions; Ear-nose-throat (ENT) infections; Gangrene; Pelvic inflammatory disease; Perforated/bleeding ulcer; Pyelonephritis; Constipation	Outcome
2.2	Chronic conditions: Angina; Asthma; Chronic obstructive pulmonary disease; Congestive heart failure; Diabetes complications; Hypertension; Iron-deficiency anaemia; Nutritional deficiencies; Gastro-oesophageal reflux disease (GORD); Osteoporosis.	Outcome
2.3	Immunisable conditions: Influenza and pneumonia; Other vaccine preventable conditions	Outcome
3.0	Comprehensive Diabetes Care: HbA1c testing; eye examinations; lipid testing; microalbuminaria screening; primary care visits	Process
4.1	1 Social indicators: Accomodation; Employment, meaningful activities and engagement; Financial support; Social contacts; Additional marginalising factors (such as ethnicity, speech differences); Safeguarding	Structure-Outcome
4.2	2 Genetic and biological indicators: Assessment of physical and mental health needs and health checks; Long Term Condition (LTC) pathways and planned reviews of need; Care Planning / health action planning; Crisis / emergency planning and hospital passports; Medication evaluation; Specialist learning disability service provision	Process
4.3	3 Communication difficulties and reduced health literacy indicators: Poor bodily awareness, reduced pain responses and communication support; Communicating health needs to others; Carers' ability to recognise expressions of needs / pain; Carers' ability to recognise and respond to emerging health problems and / or promote health literacy; Understanding Health Information and Making Choices	Structure-Process

No.	Indicators and sub-indicators	type
4.4	4 Personal behaviour and lifestyle indicators: Diet and hydration; Exercise; Weight; Substance Use; Sexual Health; Risky Behaviour / Routines	Outcome
4.5	5 Deficiencies in service quality and access indicators: Organisational barriers; Consent; Transition between services; Health screening / promotion; Primary / Secondary Care; Non health services.	Process
5.1	Enabling and partnership: Healthcare professional informs parent; trust parent as expert of the child; anticipates concerns; answers questions etc.	Process
5.2	General and specific information: Healthcare professional gives information about services in community, about child's disability, therapies, etc.	Process
5.3	Co-ordinated and comprehensive care: Healthcare professional looks at the needs of the 'whole' child (e.g. at mental, emotional and social needs), plans together with other health professionals; informs you in time about changes in care; communicates with school, ensures that family receives support.	Process
5.4	Respectful and supportive care: Healthcare professional helps parent to feel competent, provides enough time, a caring atmosphere, treats parent respectful.	Process
6.1	Individual Outcomes: Satisfaction with, and Choice and Decision-Making regarding housing, daily activities and work; Choice and Decision-Making about daily activities, housing etc.; Self-Determination: Needed and received help with daily activities/budget; Community inclusion; Work; Relationships.	Outcome
6.2	Health, Welfare, and Rights: Safety (incidence of serious injuries, mortality, support, feeling safe, victim of crime); Health (health status, received tests and screenings, health status, presence of primary care doctor); Medication; Wellness (healthy habits); Restraints; Respect/Rights (rights are respected; treated with respect by others).	Outcome-process-structure
6.3	System Performance: Service Coordination (satisfaction with received help from service coordinators); Access (capable staff; availability of transportation and support/care when needed)	Structure-process
6.4	Staff Stability: Continuity of staff presence (vacation rate, trainees, job switches)	Structure
6.5	Family Indicators: Choice and Control (Family's control/decision making about budgets; care); Family Outcomes (support for family in caring for their relative); Information and Planning (information about planning care and involvement of family); Satisfaction (of family with care for relative); Community Connections (integration of family in community); Access and Support Delivery (family reported access to and satisfaction with services and support).	Process

No.	Indicators and sub-indicators	type
7.1	1 Involvement of Children and Adults with Learning Disabilities and Their Family Carers through Self-Representation and Independent Advocacy: Involving people in planning services; in planning care across all services; Policy for access to health records; Complaints procedure; Advocacy (strategy and services are present)	Structure-Process
7.2	2 Promoting Inclusion and Wellbeing: Disability awareness (Disability Discrimination Act; Strategy; Safe Access); Transport; Policy and accessible information on Health promotion and health improvement; Health information and cultural sensitivity; Direct payments to people with ID)	Structure-Process
7.3	3 Meeting General Healthcare Needs: Assessment (of health and capacities); Care plan is present; Primary care and community services (named specialist practitioner, responsive to needs, national screening, monitoring, joint working); specific services for wheelchair and older people are present; General health and hospital services (education for healthcare professionals, advice from specialists; aware of needs; palliative care; specific illnesses)	Structure
7.4	4 Meeting Complex Healthcare Needs: Service integration (specialised & general health services); Transitions (age/service-related); Access to and availability of specialist services (Children/Adults/Complex needs/Challenging or offending behaviours/mental health problems/Autism spectrum dis./Dementia/Profound and multiple impairment/Learning disabilities and epilepsy);	Process
7.5	5 In-patient Services - Daily Life: Environment (plan and accommodation); Privacy and personalisation; Daily life (making own choices)	Process-structure
7.6	6 Planning Services and Partnership Working: Strategic health improvement and needs assessment (strategies); Database developments; Healthcare planning; Hospital closure and service reprovision; Partnership working	Structure
8.1	Demographics: Prevalence of ID in population; Living arrangements; Daily occupation; Income/socio-economic status; Life expectancy.	Outcome
8.2	Health Status: Epilepsy; Oral Health; Body mass index; Mental Health; Sensory capacities; Mobility.	Outcome
8.3	Determinants of health: Physical activity; Challenging behaviour; Psychotropic medication use	Outcome
8.4	Health Systems: Hospitalisation and contact with healthcare professionals; Health check; Health promotion; Specific training for physicians	Structure-process
9.1	Parents' Evaluation of Developmental Status: Parents have concerns (or not) about their child's learning, development or behavior.	Outcome
9.2	Comprehensive and coordinated care: The child had a personal doctor or nurse; usual source of care; parent received needed help with coordination and referrals without problems	Process

No.	Indicators and sub-indicators	type
9.3	Medical Home: a personal doctor or nurse, a usual source of care, family-centered care, care coordination if needed, no problems receiving needed referrals	Process
9.4	Elicitation of parental developmental concerns and developmental screening: Healthcare providers asked parents about concerns about child's learning, development or behavior; healthcare provider asks parents to complete an age-appropriate standardised developmental screening tool	Process
10.0	Quality care indicators of diabetes for people with ID: HbA1c checked; Lipids/cholesterol; Eye exam; Weight change; Physically active; Attended emergency department related to Diabetes Mellitus	Process-outcome
11.1	1 Shared decision making: Families of CSHCN (children with special healthcare needs) partner in decision-making at all levels and are satisfied with the services they receive	Process
11.2	2 Coordinated care: CSHCN receive coordinated, ongoing, comprehensive care within a medical home (a medical home means a source of ongoing, comprehensive, coordinated, family-centered care in the child's community)	Process
11.3	3 Adequate insurance: Families of CSHCN have adequate private and/or public insurance to pay for the services they need.	Structure
11.4	4 Screening for special healthcare needs: Children are screened early and continuously for special healthcare needs	Process
11.5	5 Community-based services: Community-based services for CSHCN are organised so families can use them easily	Structure
11.6	6 Services for transitions: Youth with special healthcare needs receive the services necessary to make transitions to all aspects of adult life, including adult healthcare, work and independence.	Process
12.1	Learning disabilities register: The contractor establishes and maintains a register of patients with learning disabilities.	Structure
12.2	Thyroid disease among people with DS: Percentage of patients on the Learning Disability register with Down's Syndrome aged 18 and over who have a record of blood TSH in the previous 15 months (excluding those who are on the thyroid disease register)	Outcome
13.1	Crucial QIs: Medication review, General health review, Restrictive practice, Excessive dose, Anti-psychotic medication, Gradual dose reduction, Dementia anti-psychotic medication.	Process

No.	Indicators and sub-indicators	type
13.2	Grade 1 QIs: Multiple medication use/polypharmacy, Anti-cholinergic medication, Anti-depressant medication, Psychotropic medications, Psychotropic/neuroleptic side effects, Dysphagia, Insomnia treatment and sleep behavior, Dementia cholinesterase inhibitors - anticholinergic medication.	Process-Outcome
13.3	Grade 2 QI: Geriatric syndromes	Outcome
13.4	Grade 3 QIs: Informational transfer, Communication, Medication reconciliation, Residential care, Pharmaceutical care/pharmacist, Non-pharmaceutical care/pharmacist, External environment, Dementia cholinesterase inhibitors, Dental-oral health, Pain, Infections, As requires 'PRN' psychotropic medications, Psychotropic medication physical side effects, Adverse drug reactions.	Process-Outcome-Structure
13.5	Grade 4 QIs: Acute behavior, Advocate, Covert administraion of medication, Inter-intra-class psychotropic multiple medication use/polypharmacy, Anti-epileptic medications, Off Label psychotropic medications, Gastro-intestinal disorders, Autism spectrum disorder.	Process-Outcome


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Quality of health care according to people with Down syndrome, their parents, and support staff – a qualitative exploration

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ABSTRACT

Background: People with Down syndrome (DS) have complex health care needs. Little is known about the quality of health care for people with DS, let alone how it is appraised by people with DS and their caregivers. This study explores the perspectives of people with DS, their parents and support staff regarding quality in health care for people with DS.

Method: We conducted semi-structured interviews with 18 people with DS and 15 parents, and focus groups with 35 support staff members (of people with DS residing in assisted living facilities) in the Netherlands.

Results: According to the participants, health care quality entails: well-coordinated health care aligned with other support and care systems, a person-centred and holistic approach, including respect, trust, and provider-patient communication adapted to the abilities of people with DS.

Conclusions: Our findings may be used to improve health care for people with DS, and provide insight into how health care could match the specific needs of people with DS.

INTRODUCTION

Down syndrome (DS) is associated with a large variety of health problems with varied severity, and consequently complex health care needs, generally involving many different health care providers (Coppus, 2017; Grieco et al., 2015; Jensen & Davis, 2013; Weijerman & De Winter, 2010). Consequentially, DS-specialised health care has evolved and in several countries DS-specific, multidisciplinary outpatient clinics – in the Netherlands referred to as 'Downteams' – have been set up (Coppus, 2017; Skotko et al., 2013; Tenenbaum et al., 2008; Weijerman & De Winter, 2010). *Paediatric* Downteams and a few *adult* Downteams are present in the Netherlands. The paediatric clinics provide team appointments including a visit to the paediatrician, physiotherapist, ENT- (ear-nose-throat) specialist and others, all on the same day. Adult teams are comprised with other specialities, related to changing needs in adulthood, and include an ID physician (a medical doctor specialised in intellectual disabillity (ID) medicine) instead of a paediatrician.

The Convention on the Rights of Persons with Disabilities advocates high-quality health care for people with disabilities, as it acknowledges the right for obtaining the highest possible level of health (UN, 2006). Strikingly, little is known about the quality of DS-specialised health care (Van den Driessen Mareeuw, et al., 2017), let alone how it is appraised by people with DS and their caregivers (Barelds, et al., 2010; Kyrkou, 2018; Minnes & Steiner, 2009). Although a number of studies have addressed the assessment of health status and quality of life of people with ID and DS (Bakker-van Gijssel et al., 2017; Graves et al., 2015; Kyrkou, 2018; Van Schrojenstein Lantman-de Valk, et al., 2007), health care quality related to people with DS has not been adequately researched (Van den Driessen Mareeuw et al., 2017). Studies that do address quality in health care for people with DS, are traditionally conducted from a medical professional's perspective (Jensen & Davis, 2013; Jespersen et al., 2018; Phelps, et al., 2012). However, it is acknowledged increasingly that insight into the patient's perspective is crucial for improving health care quality (Poitras et al., 2018; Rathert et al., 2013), answering patients' needs (Barelds et al., 2010; Phelps et al., 2012; Trebble et al., 2010), and increasing cost-effectiveness (Porter, 2010). Our aim is therefore to provide insight into the perspectives of people with DS, parents, and support staff regarding quality of health care for people with DS in the Netherlands. This includes all primary and secondary health care that people with DS may need during their lives (e.g. health care provided by paediatricians, ID physicians,

physiotherapists, dieticians etc. (within or outside Downteams), GPs). We included people with DS, their parents, and support staff (i.e. people working in assisted living facilities for people with ID and DS) in our study, for two reasons. First, it is increasingly acknowledged that patients should be seen and approached as part of a family system, in which all members collaborate with health care professionals in order to tailor health care to the needs and abilities of the patient and his/her family (Kyrkou, 2018; Rawson & Moretz, 2016). For people with DS, this system may involve parents and support staff, all playing a significant role in the lives of people with ID including DS (Mastebroek et al., 2016). Second, parents and support staff may complement people with DS' views on health care quality or may function as proxies for people with DS who are not able to verbally express themselves.

The World Health Organization (2006) identifies six dimensions of quality of care, being 1) effective (evidence based and based on needs), 2) efficient (maximising resources, avoiding waste), 3) accessible (timely, geographically reasonable, in a suitable setting), 4) acceptable/patient-centred (taking into account preferences, culture of patient), 5) equitable (same level of quality for everyone) and 6) safe (minimising risk and harm). We use these dimensions to study quality of health care for people with DS. However, we add more detail to the concept of 'patient-centeredness' by including the eight principles of patient-centred care defined by Picker (partly overlapping the WHO-dimensions): 1) respect for patient's values, preferences and expressed needs, 2) information-education, 3) coordination and integration, 4) physical comfort, 5) emotional support and alleviation of fear/anxiety, 6) involvement of family/friends, 7) continuity and transition, and 8) access (Rawson & Moretz, 2016; Singer et al., 2011).

Health (status) and (health-related) quality of life are considered to be important outcomes for assessing health care quality (Donabedian, 2005; Porter, 2010; Jespersen et al., 2018). Therefore, (health-related) quality of life is an important concept in the current study. We studied quality of life (i.e. as an outcome of quality of health care) using the eight quality of life domains of Schalock et al. (2005), because they are most frequently cited in literature and are multidimensional (Simões & Santos, 2016). They were specifically developed for people with ID and include: 1) emotional well-being, 2) interpersonal relations, 3) material well-being, 4) personal development, 5) physical well-being, 6) self-determination, 7) social inclusion, and 8) rights.

This study addressed the following research questions:

How do people with Down syndrome, their parents and their support staff define quality of health care for people with DS?

- ***What are their experiences with received health care?***
- ***How may health care influence the lives of people with DS?***

METHOD

This article uses the 'Consolidated criteria for reporting qualitative research' (COREQ), a checklist for qualitative research that "aims to promote complete and transparent reporting (…) and indirectly improves rigor, comprehensiveness and credibility" (Tong et al., 2007).

Study design and research team

The study has a qualitative design, using a constructivist approach, which acknowledges that people may have different perceptions of reality as a result of different experiences or (social) interactions (Tavakol & Sandars, 2014). We conducted semi-structured interviews with people with DS and with parents of people with DS, and focus groups with support staff. The study was approved by the Ethical Committee of the School of Social and Behavioral Sciences of Tilburg University (Tilburg, The Netherlands) on August 21, 2016 (no. EC-2016.21).

The research team consisted of a paediatrician with expertise in integrated care for people with DS (professor) and data driven research (EV), an expert in health services research (professor) and quality measurement (DD), an ID physician and epidemiologist with expertise in DS (senior researcher) (AC), and a health scientist (master's level training) with expertise in public health and qualitative research involving people with ID (FDM).

Participants

Purposive sampling was used to collect as many experiences, opinions and ideas about quality of health care for people with DS as possible, by including participants with DS who differed in terms of age, gender, living situation, geographical location, and medical problems. They had to be able to take part in an interview; we therefore included people ≥12 years with mild to moderate

intellectual disability. We also strived for diversity regarding the people they care for and regarding their personal characteristics within the groups of parents and members of the support staff. This included parents and support staff of people with DS with a larger age-range (also younger than 12), and of people with DS with more severe intellectual disability, than the group of participants with DS. Support staff had to be involved in providing health care for at least one person with DS (e.g. join patient consultations, prepare consultations with patient).

Participants were recruited through the Dutch DS Association, through service organisations for people with ID, as well as by means of the network of the authors. Interested parents contacted FDM by e-mail or telephone after which they received an information letter and an informed consent form for themselves and/or for their child with DS (in easy-to-read format). Service providers were approached by using publicly available contact details or via a contact person out of the professional network of the authors. Five (including three in the authors' networks) of 36 contacted service providers agreed to participate. Service providers mentioned the following reasons for not participating: they "did not have time to participate", "did not see the relevance of the study", "did not agree with the focus merely on DS (instead of on people with ID)", or "thought the effort for clients / staff would be too great". We obtained contact details of (coordinating) support staff members working at assisted living facilities with 24h or floating support, or at daily activity centres for people with ID from the five participating service providers. AC worked at one of the participating service providers, and identified eligible participants, as a result of which she knew several of the participating support staff members. AC was unaware of who eventually participated, nor did she know which data originated from which support staff member. There were no other relationships between the authors and the participants prior to the study. All support staff members whose contact details were obtained, received information letters (for people with DS, parents and support staff) and identified eligible persons, and they were asked whether they wanted to participate themselves. They provided us with contact details of parents of people with DS, and/or arranged interviews with people with DS, and/or arranged focus groups with support staff. The contact person of one of the non-participating service providers acknowledged the relevance of the study and asked her relative with DS (+parents) to participate. An additional potential participant (parent of a person with DS) was identified during a site-visit by FDM.

Participants and/or their legal representatives gave informed consent. Interviews and focus groups were planned after informed consent forms were received (by (e-)mail). 18 people with DS and 15 parents or parent couples were interviewed. Two parents initially agreed to participate, but one withdrew because of sudden illness of her child, and with one contact was lost. In total 34 support staff members from the five different service providers participated in five focus groups, of respectively two, seven, nine, and twelve participants. One support staff member was unable to attend the focus groups and was therefore interviewed individually. In one case, the person with DS, his parents as well as his support staff participated in the study. In 11 cases, both people with DS and their parent(s) participated. In six cases both people with DS and their support staff participated. Characteristics of participants are shown in Table 3.1.

Table 3.1 Participant characteristics

	Persons with DS (n=18)	**Parents/parent couples (n=15)**	**Support staff (n=35, supervising a total of 25 persons with DS)**
Age (y) *Mean [range]*	31,7 [13-54]	57,3 [37-79]	39,8 [21-59]
Gender *Female; Male*	10; 8	14; 6 (5 parent couples, 9 mothers, 1 father)	27; 8
Geographical location within the Netherlands[a]			
South:	10	5	27
Other:	8	10	8
Living situation:		n/a	n/a
Family living:	4		
Living with floating support (during mornings and evenings):	11		
Living with (almost[b]*) 24h support*	3		
Level of ID[c]		n/a	n/a
Borderline (IQ 70-85):	2		
Mild (IQ 50-70):	8		
Moderate (IQ 35-49):	7		
Severe (IQ 20-34)[d]*:*	1		
Health problems[c]		n/a	n/a
Mentioned in number (and percentage) of interviews[e]			
Vision problems:	13/18 (72%)		
Foot / walking problems:	13/18 (72%)		
Overweight:	10/18 (56%)		
Thyroid dysfunction:	6/18 (33%)		
Heart problems:	5/18 (28%)		
Sleeping problems / apnoea:	4/18 (22%)		
Hearing problems:	3/18 (17%)		
Coeliac disease:	2/18 (11%)		
Psychological problems:	2/18 (11%)		

Table 3.1 continued

	Persons with DS (n=18)	Parents/parent couples (n=15)	Support staff (n=35, supervising a total of 25 persons with DS)
Living situation of child/client(s) with DS)	n/a		
Family living:		11	
Living with floating support (during mornings and evenings):		3	16
Living with (almost[b]) 24h support:		1	9
Level of ID of child/client(s) with DS[c]	n/a		
Borderline (IQ 70-85):		3	
Mild (IQ 50-70):		4	8
Moderate (IQ 35-49):		6	14
Severe (IQ 20-34):		1	1
Not yet assessed (too young):		1	
Dementia:			2
Health problems of child/client(s) with DS[c]	n/a		
Mentioned in number (and percentage) of total number of interviews or focus groups[e]			
Skin problems:		12/15 (80%)	6/6 (100%)
Vision problems:		10/15 (67%)	2/6 (33%)
Foot / walking problems:		-	4/6 (67%)
Dementia:		8/15 (53%)	4/6 (67%)
Overweight:		7/15 (47%)	3/6 (50%)
Thyroid dysfunction:		7/15 (47%)	2/6 (33%)
Heart problems:		4/15 (27%)	3/6 (50%)
Sleeping problems / apnoea:		2/15 (13%)	2/6 (33%)
Hearing problems:		2/15 (13%)	3/6 (50%)
Psychological problems:		2/15 (13%)	-
Functional decline:		-	3/6 (50%)
Behavioural problems:		-	3/6 (50%)
Age of child/client(s) with DS *Mean [range]*	n/a	24,1 [2-43]	44,3 [24-63]
Gender of child/client(s) with DS	n/a		
Female; Male		7; 8	13; 12
Professional experience with people with DS (y):	n/a	n/a	
<5			5
5-10			12
>10			18

a The authors are based in the south of the Netherlands, which resulted in more cooperating service providers in the south. (See: "Participant selection and recruitment")

b Some locations had an overnight surveying system, without support staff being physically present.

c Parents or support staff provided data on most recent IQ/development test (in the Netherlands, this generally includes an IQ-test and a performance test) and on basic physical health. Information on physical health was also obtained during the interviews / focus groups.

d One participant wanted to join despite the fact that this person had a severe intellectual disability.

e if mentioned in 2 or more interviews focus groups

In both the interviews and focus groups, data saturation occurred: additional interviews / focus groups did not yield new relevant information (Tong et al., 2007).

Setting

Participants with DS chose the time and venue of the interview: at their home, their parents' home, or at their work. Participants could invite someone else to join the interview, for emotional and/or verbal support. Eleven participants invited their parent(s), five invited a support staff member. As stress-diminishing measure the interview could be split in two: the first part to get acquainted with the interviewer and with "participating in an interview", the second part focussed on the content (quality of health care and life). However, all but one participant preferred one single interview, due to time constraints or expected possible burden of two interviews. The interviewer adapted the interview to the participant's abilities (for example, adjustments were made with regard to talking pace, length of sentences, words used and extent to which supporting visual materials were used). The interviews with people with DS lasted 30 to 75 minutes.

Parents were also free to choose the time and venue of the interview: at home, by telephone, at their child's home (assisted living facility) or work. In the latter two cases, their child with DS was interviewed before or after the parents' interview. The interviews with parents lasted 30 to 105 minutes.
The focus groups with support staff and the interview with one support staff member took place in meeting rooms of the service providers. Three focus groups were attended by support staff members from one service provider, the other two focus groups had participants from two organisations. Travelling costs to the venue where the focus groups took place were reimbursed. The focus groups took about 30 minutes to two hours (depending on time available by participating support staff) and the single interview lasted 50 minutes.

The interviews with people with DS and with parents took place during the period from April until September 2017, the focus groups and interview with support staff in December 2017 and January 2018. All interviews and focus groups were conducted, respectively convened by FDM.

Topics discussed

An interview or focus group guide was composed for each specific group of participants (people with DS, parents, and support staff). The different guides included similar topics based upon the eight domains of quality of life as formulated by Schalock (2005) and patients' experiences (in this case of people with DS, together with their parents and/or support staff) during their journey along health care, the 'patient journey' (Trebble et al., 2010). The 'patient journey' is defined as the "series of consecutive events or steps" related to a treatment or condition (Trebble et al., 2010). Additionally, the guide contained an introduction section, providing participants with information about the study and its aims. It explained the course of the interview or focus group, and put participants at ease. Participants were also allowed to add topics they thought were important. Although the content of the guides for each group of participants was similar, the way in which the topics were discussed differed in terms of detail and order of topics, in order to match the participants' (cognitive) abilities, backgrounds, and experiences. The interview guide for interviews with people with DS included pictures (of e.g. health care providers) and pictograms (e.g. representing abstract concepts like "sad" or "bored"). A draft of the interview guide for people with DS was discussed (and adapted accordingly) with other researchers with experience in interviewing people with mild to moderate ID. A summary of the interview guides and some example questions are presented in Appendix 3-I.

Data processing and analysis

All interviews and focus groups were audio-taped, after receiving all participants' permission, and pseudonymisely transcribed. Pseudonymised transcripts were sent to the participants in order for them to check the transcripts and make adjustments if desired. Due to limited literacy skills, participants with DS received a verbal summary of the interview at the end of the interview, after which they could refine or add things. Transcripts and personal data were stored in a protected digital environment.

Data analysis was based on the Framework Analysis Method (Gale et al., 2013), see Table 3.2. All authors were involved in data-analysis (including coding). To maximise objective analysis, one third of the transcripts was double coded by two authors (by FDM and AC, DD, and EV, respectively). Data was managed using the software package Atlas.ti 8 for Windows.

Table 3.2. Data analysis consisting of three successive steps, based on the Framework Analysis Method (Gale et al., 2013).

Step	Description
1. Coding	Reading first few transcripts and labelling text fragments with codes reflecting relevant/interesting information. This was done using a combination of inductive (open) and deductive (using predefined codes) coding (Gale et al., 2013), which ensured that important themes in the data were not missed and enabled structuring the complex data. Predefined codes derived from theory: Quality of life domains (Schalock et al., 2005), quality of care dimensions (WHO, 2006), principles of patient centred care (Rawson & Moretz, 2016; Singer et al., 2011).
2. Constructing and applying analytical framework	Codes were grouped into themes indicating interrelatedness and variety of the topics covered by the transcripts. The framework (see Appendix 3-II) was then applied to other transcripts. This was done in three iterations.
3. Charting data	Charting the data in a framework matrix (see Appendix 3-III), allowing interpretation.

RESULTS

In describing the results, we use 'participants with DS / people with DS' or 'parents' if we mean (parents of) people with DS of all ages. Findings pertaining to a specific age group are indicated by 'child', 'adult' or other age indication. The findings originating from support staff always pertain to adults with DS.

Life and health

Participants with DS reported that they were happy, and satisfied with their living situation and daily activities, although others felt lonely or reported being bullied because of having DS. They either liked to have DS, or did not like it, or did not think they had it. Both positive and negative issues were confirmed by parents and support staff, although support staff did not address the topic "what about having DS".

Participants with DS were well informed about their health (problems) and considered themselves quite healthy, although they suffered from many different health problems (e.g. hearing/vision/skin problems, sleep apnoea, psychological problems, celiac disease, thyroid dysfunction, and a history of heart problems or leukaemia), reflecting the specific health profile of people with DS (Grieco et al., 2015; Kinnear et al., 2018). Interviewed parents presented a similar picture: *"She's never ill, but there's always something the matter with her."* (mother (55yrs) of woman with DS (23yrs)). Parents either indicated that health problems were managed well,

generally resulting in a low burden, or experienced difficulties with managing the complex health care needs. Support staff too considered people with DS as being quite healthy, but also mentioned a lot of health problems their clients with DS suffered from, including physical and mental decline and dementia (Coppus et al, 2017).

Health care utilisation and 'Downteams'

According to participants with DS, parents and support staff, people with DS received, or had received, care by a large variety of health care providers. Roughly spoken, the paediatrician and speech therapist were visited during childhood; ID-physician, general practitioner, and dietician during adulthood; physiotherapist, internist, ophthalmologist, ENT-specialist, and psychologist during childhood and adulthood.

Participants with DS and their parents were visiting or had visited a *paediatric* Downteam. An important reason mentioned by parents for visiting a paediatric Downteam is that multiple specialists can be visited in one day, which they think is efficient and provides them with good information and advice. Parents also explained that the team offered regular health checks and screenings allowing for timely detection of health problems, preventing problems worsening, and identification or ruling out of physical causes of behavioural problems. The latter was deemed especially important for people with DS who are less able to display pain or other symptoms of disease. The reasons mentioned by parents are in accordance to the reasons mentioned in literature supporting the relevance of such teams. It is argued that Downteams are crucial in monitoring health, discovering hidden health problems, and preventing complications (Skotko et al., 2013; Tenenbaum et al., 2008; Weijerman & De Winter, 2010).

Parents who were positive about the paediatric Downteam preferred to have more influence on the type and sequence of health care providers scheduled for their child. Other parents, not visiting the teams (any more), thought that a visit to a Downteam would lead to too many referrals, or deemed a regular check-up unnecessary, arguing that they did not want to medicalise their son/daughter and that they would visit a doctor when needed. Other reasons for not visiting the teams were unawareness about the existence of the teams, or the absence of one nearby.

Whether adult participants with DS went to *adult* Downteams, depended on the awareness among people with DS, parents and support staff about the existence of such teams and on the teams' geographical proximity. Parents and support staff thought such teams would be very useful. According to parents, a barrier for visiting adult Downteams is due to the fact that some of them are located at a venue of an institution for people with ID instead of, for instance, in a general hospital and/or within the community.

Role of parents and support staff

Participants with DS, parents and support staff reported that people with DS generally needed support deciding about visiting a doctor, making appointments with health care professionals, communicating during consultations, and sharing health or treatment information with (other) health care professionals, support staff, parents or other relatives. This is in line with literature on adults with ID in primary care (Mastebroek et al., 2016). When people with DS were living with their parents, parents offered this support. People with DS living in an assisted living facility, received this support from support staff and/or parents/other relatives. There were also adult participants with DS who reported that they visited nearby health care providers on their own. Parents and support staff stressed that especially in such cases, it is important that health care professionals share information about treatment or diagnoses with the caregivers of their patient with DS. Support staff and parents indicated they did not always agree about needed health care for their child/client with DS. Support staff revealed that parents' attitudes towards the health care needed for their son/daughter with DS ranged from being quite indifferent, to over-demanding. This sometimes led to discussions between parents and support staff about what is best for the person with DS. Parents expressed worries such as 'Does support staff notice symptoms of my son/daughter in time?' and 'What will happen with my son/daughter when I die?', especially when their child would soon be leaving home, or when parents were old. Parents and support staff agreed that support staff did not have a high level of (DS-specific) medical knowledge, which is consistent with the literature (Mastebroek et al., 2016).

Perceived health care quality

Generally, participants with DS, parents and support staff qualified health care for people with DS as good, although less positive stories also were heard

regarding health care for (especially) adult people with DS, including rude health care providers, health problems that were not taken seriously, difficulties in getting an appointment, and inpatients who were neglected because staff was unaware of (eating) (dis)abilities. According to participants with DS, health professionals are 'good' when they cure their health problem. Parents and support staff also considered general (not DS-specific) medical expertise of health care professionals as important, or took this for granted. Parents and support staff mentioned that expertise on *DS-specific* common health problems and symptoms was an important - although not always present - element of health care quality, especially regarding adult people with DS, for whom Downteams are scarce in the Netherlands. Parents also explained that good health care nearby, at least within the region they lived in, was important due to time constraints. They however understood that it is unrealistic to expect all health care professionals to be DS-experts, or specialist health care to be 'around the corner'. Other parents did not mind travelling further for good health care. Parents of especially adults with DS also explained that DS-specific expertise is not always needed, as long as professionals know where to find expertise, where to refer to, and adapt treatment to the personal needs and abilities of their son/daughter with DS. Additionally, parents indicated the importance of effective and efficient care: *"You just want to be helped effectively, it shouldn't cost too much time. […] 'cause a child with DS costs a lot of time and energy. Doctors should realise that"* (Mother (49yrs) of a boy with DS (13yrs)). Similar time and energy constraints are reflected in literature (Phelps et al., 2012; Povee et al., 2012).

Holistic approach and benefit-burden balance

Participants with DS, parents and support staff indicated that health care should be oriented around the needs, preferences, and abilities of people with DS. Parents and support staff underlined that health care professionals should apply a holistic view regarding their patients with DS, which they defined as integrating different health problems of their son/daughter/client, but also connecting health (care) to other dimensions of life, such as personality, personal goals, lifestyle, physical and social environment and life phase. A holistic approach was also advocated by (parents of) people with ID in other studies (Minnes & Steiner, 2009; Kyrkou, 2014). According to parents and support staff, applying a holistic approach also means that health care professionals determine together with their clients with DS and their caregivers which care is actually needed to improve the client's well-

being. They explained that, compared to the general population, the burden of treatment may be much more significant than the benefit for a person with DS. All participant groups gave a number of examples of health problems with a large impact on life (high benefit if treated), especially concerning adults with DS: sleep apnoea (impaired daily functioning and behaviour, not always detected), communication problems (impedes emotional expression and social interaction), walking problems (influences functioning and independence, cause often unidentified). The following quote is an example of how burden and benefits are taken into consideration when weighing health care options: *"We explored that [treatment] option, but it's quite an intervention, which can be painful too. (…) finally we decided not to do anything as long as he [son] does not indicate pain or move differently."* (father (54yrs) of a boy with DS (14yrs)). Goodman and Brixner (2013) confirm the importance of considering the impact of a treatment on quality of life in people with DS.

Adapted communication, trust and respect

Specific communication difficulties, such as language processing or hearing problems, commonly present among people with DS (Grieco et al., 2015) may hinder communication between health care professionals and their patients with DS. Adult participants with DS argued that health care professionals should communicate well with the person with DS: talking slowly, not using complex words, and explaining what happens, for example during dental treatment or small surgery, or explaining step by step what is going to happen, for example during surgery. Furthermore, they preferred professionals whom they had been knowing for a longer period of time, and with whom they built a trust relationship. This would create a comfortable atmosphere in which talking about health problems is easier: *"If they know me well, then I talk more. (…) Because then I know I can trust that person."* (woman with DS (54)). Other qualities mentioned by participants with DS were: being kind and reassuring, asking about other – not medical – things, making jokes, and taking time to listen. Parents and support staff acknowledged the relevance of these communicational and relational issues. They added that adapting communication to the inner world of people with DS is important, that using pictures may be helpful, and that talking to, instead of about, a person with DS is key. They considered this a matter of respect that contributed to a feeling of 'being seen and heard': *"quality of care is quality for the patient, looking the patient in the eyes, listening to his story, not being focused only on a diagnosis, but just asking*

'how are you, what's the matter, can you tell me more?' ". (father (54yrs) of a boy with DS (14yrs)). Similar issues were found in studies on health care for people with ID (including DS) (Mastebroek et al., 2016; Miller et al., 2009). However, in people with DS, these issues may need even more attention, because communication difficulties are prominent among people with DS and they may have different cognitive and behavioural profiles, including different pain representation, compared to people with ID (Grieco et al., 2015; Kyrkou, 2014).

Complexity of (health) care

Although participants considered health care quality to be important, especially parents explained that health care was just one of many services to be managed. Parents, mainly of younger children with DS, even argued that arranging health care was easy, and that arranging developmental or other support was more challenging: *"The medical care around these downers [people with DS] is fine, that's not the biggest problem, it's the rest, developmental and educational problems. I'm also involved in a Downteam as a professional and almost all parents have got these problems, like we do."* (mother (49yrs) of a boy with DS (13yrs)). Especially those parents, but also parents of older/adult children, experienced stress caused by problems in finding and (financially) arranging (developmental) support, dealing with related paperwork and regulations, and with the complexity of organisations involved. Additionally, parents of especially younger children with DS reported problems with integrating health care with other services, for example making sure that educational support at school matches the methods used by the speech therapist and vice versa, or with their daily family schedule, especially when parents had more children: *"I just want to integrate it in our life, in how we do things. […] I don't want the speech therapist to be annoyed because I did not do my 'homework' with him [son with DS]".* (mother (57yrs) about her son (man with DS (26yrs)) during childhood). Other parents did report problems in arranging medical care in addition to arranging all other services: *"going to the podo-therapist, orthopaedist, dentist, ophthalmologist, physiotherapist every week; and that's only the medical part. Then maintaining her room, repairing her clothes. And the conversations with the service provider, the ID-physician, and what else? The yearly evaluation of her personal support plan, next month a meeting about her depression, and next week to the hospital. […] It's just the combination of it all.[…] and it's always fighting for everything, always. And everything changes, different regulations, and all the paper work…"* (mother (63yrs) of a woman with DS (28yrs)). Minnes & Steiner (2009) also

observed this "stress in dealing with the health care system and in negotiating relationships with practitioners".

There were also parents of people with DS in the childhood age who had created a well-coordinated team of care and support around their son/daughter, mostly supported by local authorities or benefits. They argued that their own managing and coordination skills were crucial in creating such networks: *"If you're not capable enough as a parent, having cognitive skills or financial capacity, then your child [with DS] does not receive the right care, and suitable education is an illusion".* (mother (37yrs) of a girl with DS (7yrs)). Povee et al. (2012) acknowledge this diversity in coping with organisational challenges and argue that for families with limited advocacy skills it is hard to obtain the needed services.

A need for coordination

According to parents and support staff, collaboration and good communication between all the different professionals involved are important elements of health care quality. This notion is supported by literature on the topic (Miller et al, 2009; Kyrkou, 2014). Participants with DS did not mention such issues. Furthermore, parents indicated that they would like to have more information on where to find the right health care provider(s) for their son/daughter. They argued that ideally, a professional should be available who acquires an overview of the complexity of different health problems of their child with DS, coordinates, and helps finding needed health care: *"he [son with DS] has a lot of different unexplained health problems. Then it's nice to have a trust relationship with someone […] a coordinating person, that would be nice."* (mother (57yrs) of a man with DS (25yrs)). According to parents, this professional should also connect with actors outside health care, for example school, daily activity centre, social services. This coordinating role was not allocated to a specific professional, but could be, or was, fulfilled by a paediatrician, GP, ID physician, or representative of a service provider.

Parents and support staff furthermore expressed the need for continuity in care providers. They experienced that many changes in care providers impeded good coordination and the establishment of the above-mentioned necessary trust relationship. Parents and support staff stressed the importance of good coordination in the case of transition from paediatric towards adult health care, which is complicated by the fact that paediatric Downteams are not accessible

anymore and adult Downteams are scarce: "*first the paediatrician takes this role, but as soon as he turns 18, they say: "sorry, we cannot do it anymore", there's no one who takes over.*" (mother (57yrs) of a man with DS (25yrs)). The importance of smooth transitions, good coordination, and continuity is confirmed in literature (Dyke et al., 2013; Kyrkou, 2014; Miller et al., 2009; Woodward et al., 2012).

DISCUSSION

We explored what people with DS and their representatives (parents and support staff) consider to be health care quality and how this may impact people with DS' quality of life. In summary, people with DS stressed the importance of health care professionals who cure the health problem, communicate clearly, build a trust relationship, and also pay attention to other things in life that are not necessarily related to the health problem. Parents also underlined the importance of a holistic approach and added that coordination of all services involved, including services outside the medical domain, is an important element of health care quality. Support staff complemented that for people with DS respectful treatment and creating a feeling of 'being seen and heard' are also a key for quality of health care. Parents and support staff indicated furthermore that the type of services/ professionals involved differs for each person with DS and that coordination of the transition from paediatric towards adult health care needs special attention.

Our findings are similar to the findings of studies on health care quality in general (not DS-specific) (Morgan & Yoder, 2012; Di Blasi et al., 2001). However, it is argued that compared to the general population, and to people with ID, people with DS have a specific combination of health (and other) problems (Grieco et al., 2015; Kinnear et al., 2018; Kyrkou, 2014; Minnes & Steiner, 2009; Weijerman & De Winter, 2010), which demands specific health care (provision) (Goodman & Brixner, 2013; Grieco et al., 2015; Kinnear et al., 2018; Skotko et al., 2013).

Our study provides insight into these DS-specific health care requirements. First, according to participating parents, benefits and burden of a treatment may be different for people with DS compared to the general population. This means that health care professionals should determine the best outcome (low burden, high benefit), by considering DS-specific conditions, and acknowledging the living /

family situation of people with DS and stress experienced by families. Second, the specific profile of people with DS requires adapted professional-patient interaction. Therefore, health care professionals should adapt their communication to the abilities of their patients with DS, and build a trust relationship. This may include dealing with hearing/speaking problems, text processing time, different pain presentation, and specific behaviour. Determining best outcomes and adapting communication may require extra effort from health care professionals. However, research in the general population has shown that applying such a person-centred approach does *not* require extra time from professionals and leads to more efficient care (Levinson et al., 2000; Stewart et al., 2000). Third, the care and support system is complex and includes a specific combination of a large number of health care and other professionals. Coordinating this complex system around children and adults with DS requires good management skills of parents / other carers of people with DS. Hence, coordination between the different professionals within and outside health care may be extra important. Downteams are helpful in the coordination of care, but generally do not, or only to a small extent, cover coordination with professionals outside health care. There were parents in our study who had a (non-medical) professional who coordinated the care for their child, which they considered to be very helpful. Such a 'patient navigator' has shown its effectiveness in care for people with special/complex health care needs (Dimitropoulos et al., 2019).

Altogether, this study shows that person-centeredness (determining the best outcome, taking into account the patient's specific needs and situation, using adapted communication, being respectful) and coordination are especially crucial in health care for people with DS, in both children and adults. However, person-centred care is not standard practice, health care is traditionally orientated around curing separate conditions instead of addressing the total picture, and care is organised within separate silos (Kinnear et al., 2018; Valentijn et al., 2013; Wiering et al., 2016), which is also seen in our results. Attention is increasingly directed towards integrated care models as an answer to fragmented care, lacking person-centeredness (González-Ortiz, et al., 2018). Although studies investigating the effect of integrated care models on outcomes are scarce, integrated care is considered promising in health care for people with complex needs and/or chronic disease (Busetto, et al., 2016; Gonzáles-Ortiz et al, 2018; Van Duijn et al., 2018). In integrated care, coordination of (medical and social) care, around people's needs

(person-centred), is crucial–(González-Ortiz et al., 2018). The user-led definition illustrates the meaning of integrated care from a patient's perspective: *"My care is planned with people who work together to understand me and my carer(s), put me in control, coordinate and deliver services to achieve my best outcomes"* (WHO Europe, 2016). Considering these definitions and the findings of our study, an integrated care model would be recommendable for health care for people with DS. Implementing an integrated care approach requires changes in different dimensions in the care system. Alignment of policies and rules, establishment of collaboration networks between organisations and professionals, and shared values and aims are necessary to achieve this (Valentijn et al., 2013). Such efforts are worthwhile as they lead to more efficient and effective health care (Porter, 2010; Valentijn et al., 2013).

Strengths and limitations

This study's strength is that it investigated health care quality through the eyes of people with DS and their caregivers. This perspective is crucial in determining what person-centred care for people with DS really should be, which is a requirement for improving health care quality. Another strength is that we included (parents and support staff of) children and adults with DS. The findings are therefore sensitive to health care needs in different life stages.

A limitation of the study is that selection bias may have occurred in three ways. Firstly, participation was voluntary, which may have resulted in highly motivated participants, in combination with participants who are extremely unsatisfied about health care. Secondly, people with DS with limited literacy skills or cognitive abilities could not take part in the interviews. Thirdly, about half of the participants were located in the southern part of the Netherlands. This potential bias was minimised by including people from different backgrounds (regarding age, gender, living situation), and by interviewing parents and support staff representing people with DS with lower cognitive abilities. Furthermore, all kinds of health problems known to be common in DS were present among the participants. The group of participants reflects the diversity of the DS population in this respect. Another limitation is related to the following: although the study design required open interview questions, it was not always possible to pose open questions to the participants with DS, due to their cognitive abilities. The potential effect of this limitation was curtailed by posing additional questions, similar questions in

different words, and by using visual materials, which encouraged participants with DS to express their own opinion.

Conclusion

This study contributes to existing knowledge on quality of health care for people with DS and provides insight into what are, according to people with DS, parents and support staff, crucial elements in health care. Our findings may be used to improve health care for people with DS and may also contribute to well-being of people with DS, since a higher level of health care quality contributes to better functioning (Phelps et al., 2012). Health care for people with DS should focus (more) explicitly on person-centeredness in order to answer to the specific health care needs of people with DS. An integrated care model could be helpful in reframing health care for people with DS. Future research should investigate health care providers' views on applying such approach and on quality in health care in general, in order to identify possibilities for improvement and implementation of principles of integrated care.

APPENDICES

Appendix 3-I

Overview of interview / focus group guides and example questions

Topic	Examples of questions in:		
	Interview guide for people with DS	**Interview guide for parents**	**Focus group guide for support staff**
Introduction	*Everything you tell me will remain secret. I will not tell those things to other people.*	*All information that comes up during this interview will be handled discretely.*	*All information that comes up during this meeting will be handled discretely.*
Emotional well-being	*How do you feel?*	*How can you tell your son/ daughter is happy?*	*How can you tell your client(s) with DS is/are happy?*
Interpersonal relations	*Which people are important to you? Why?*	*Which people are important to your son/daughter? Why?*	*Which people are important to your client with DS? Why?*
Material well-being	*What do you think about where you live?*	*What does your son/ daughter think about where he/she lives? And what do you think about that?*	*What does your client(s) with DS think about the living facility?*
Personal development	*What school did / do you go to? What would you like to learn?*	*What school did / does your son/daughter go to? Does he/she have things he/ she wants to achieve?*	*Do(es) your client(s) have things he/she wants to achieve?*
Physical well-being	*What do you think is healthy? Are you healthy?*	*How about the physical health of your son/daughter?*	*How about the physical health of your client(s) with DS?*
Self-determination	*What are you going to do this weekend? Who decided about this?*	*How independent is your son/daughter?*	*How independent is your client with DS?*
Social inclusion	*Do you ever go out, to the movies, for a drink with someone, etc? With whom?*	*In what social activities does your son/daughter participate?*	*In what social activities does your client with DS participate?*
Rights	*What do you think about joining in? Do you ever feel you may not or cannot join in? What happened?*	*Do you think your son/ daughter 'fits in'? Please give an example.*	*Do you think your client with DS 'fits in'? Please give an example.*
Patient journey	*Did you ever visit a: physiotherapist, general practitioner, etc.*	*Which health care providers did your son/daughter visit in his/her life?*	*Please mention one health care provider your client(s) with DS have visited in the last year. (one support staff member after the other, until no new providers are mentioned)*
Health care quality	*Who is the best doctor you've ever had? Can you tell me why?*	*What is the first thing that comes in mind when you think about quality in health care for people with DS?*	*What is the first thing that comes in mind when you think about quality in health care for people with DS?*
Other	*Are there other things you would like to tell me?*	*Are there things you would like to add, which you think are important regarding quality of life or quality of care of people with DS?*	*Are there things you would like to add, which you think are important regarding quality of life or quality of care of people with DS?*

Appendix 3-II

Analytical framework used in analysis, including codes and information on whether codes were derived from data or literature.

Code	Derived from
Quality of care: effective	Literature (WHO, 2006)
Quality of care: efficient	Literature (WHO, 2006)
Quality of care: equity	Literature (WHO, 2006)
Quality of care: safe	Literature (WHO, 2006)
Quality of care: person-centred *Sub-codes:* - Person-centred: Patient preferences and values - Person-centred: Information, communication and education - Person-centred: Physical comfort - Person-centred: Emotional support and alleviation of fear/ anxiety - Person-centred: Involvement of family and friends	Literature (WHO, 2006) - *Literature (Rawson & Moretz, 2016)*
Quality of care: accessible	Literature (Rawson & Moretz, 2016; WHO, 2006)
Dealing with complexity of care system *Sub-codes:* - Complexity care system: shared responsibilities - Complexity care system: coordination and integration - Complexity care system: continuity and transition	 - Literature (Singer et al., 2011) - Literature (Rawson & Moretz, 2016) - Literature (Rawson & Moretz, 2016)
Health care utilisation, support and aids (patient journey)	Data & literature (Trebble et al., 2010)
Information about health care, support and DS	Data
Health literacy and lifestyle	Data
Quality of life: Physical and mental health	Literature (Schalock et al., 2005)
Quality of life: Autonomy, self-control, self-perception	Literature (Schalock et al., 2005)
Quality of life: Personal development	Literature (Schalock et al., 2005)
Quality of life: Activities	Data
Quality of life: Participation and acceptation by society	Literature (Schalock et al., 2005)
Quality of life: Social environment	Literature (Schalock et al., 2005)
Impact DS on others	Data
Influence quality of care on quality of life	Data

Appendix 3-III

Framework matrix

The framework matrix was created during the analysis of the transcripts of interviews and focus groups with people with DS, parents, and support staff. The matrix provides an overview of the complex data by showing the main findings per participant group. It enables comparison between the three groups and interpretation of the data (see right column).

Theme	Said by:			Interpretation by authors
	People with DS	***Parents***	***Support staff***	
Physical health	- Generally healthy - Various health problems mentioned	Idem	Idem	- People with DS are well informed about own health - People with DS are quite healthy, most health problems are controlled well - Mentioned health problems are known to be common among people with DS.
Mental health	- Mentioned emotions: Happy, joyful, afraid of several things, sad (especially about deceased loved ones), bored, feeling lonely. - Thoughts about DS: "I don't have it", "I don't want to have it", "It's quite ok to have it".	- All kinds of emotions were mentioned to be present among their children with DS. - Children don't want to have DS, or are frustrated about having a disability.	- All kinds of emotions were mentioned to be present among their clients with DS. - Idols and deceased loved ones often play import role.	- No specific emotional issues. - Deceased loved ones are important. - Large differences in how people with DS perceive their condition(s).
Autonomy and self-perception	- Free to choose activities / work, place to live, freedom of choice is sometimes limited by available transportation. - Some were frustrated about being 'different'. - They show achievements, independence (e.g. having own apartment / having job), and that they are like others.	- Their children have freedom of choice regarding activities / work, place to live, but also in health care. However, parents try to influence this (for the best interest of their child). - People with DS have the right to have privacy. - Many of their children with DS have problems with being different and self-esteem. - Try hard to improve (feeling of) independence of their children with DS.	- Think autonomy is very important. - Try hard to improve (feeling of) independence of their clients with DS.	- Freedom of choice is important for well-being, also in health care. - People with DS want to be just like others (and are sometimes frustrated that they're not). Parents and support staff try to create 'normal lives'.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Daily activities	- School, work, day activities. - Leisure time: various activities (acting, sports, handicrafts, computer, going out, domestic tasks, going on holiday)	Idem Whether an activity is considered as nice by their son/daughter with DS is largely dependent on who joins (support staff / other participants)	Idem	People with DS have busy lives.
Personal development	- All attended school, most did internships, some attended courses. - Most had desires or personal goals for the future.	- It's difficult to find the right school and aligning education with care/support. - Some children go to regular schools (with extra support), others to specialised schools. - Switch to special education was a relief. - "Specialised school didn't teach him//her anything". - Some parents expect too much from their children with DS. - At a certain age - quite young (30-40) -development stops, deterioration starts. - Parents report cases of both over- and underestimation of the (cognitive) abilities of their child with DS.	- Some parents expect too much of their children with DS. - Support staff offers more room for making mistakes (hence for learning) than parents do. - At a certain age, development stops, deterioration starts. - Support staff report cases of both over- and underestimation of the (cognitive) abilities of their client(s) with DS.	- People with DS have goals in life and learned a lot. - Parents are surprised about stagnation of development and deterioration at relatively young age. - Expectations of parents may be too high.
Aging		- Loosing willingness to do things at rather young age (30-40y) - Occurrence of dementia	Becoming more rigid	Becoming older may cause problems.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Participation in - and acceptance by - society	- Some report being bullied - Not always able to participate because of disability (e.g. communication problems)	- Some are being bullied or not accepted as they are. - Society is too complex for them. - Media show an unrealistic (too positive) picture of people with DS, regarding their abilities and independence. - Lack of transportation and communication problems hinder participation. - Parents do a lot in stimulating participation.	- Bullied by housemates who don't want to be seen with a person with DS. - Society is too complex for them. - Media show an unrealistic (too positive) picture of people with DS, regarding their abilities and independence and "all people with DS are musical".	- Participation is an issue / problem. Large variety in sample. - Communication skills seem to play an important role in this.
Social environment	- (sometimes deceased) parents, siblings, other family and support staff are important in their lives. - Also mentioned as important: boy/girlfriend, friends, housemates, colleagues (especially those without DS/ID). - Some are lonely, others are always busy.	- The answer to the question "who is important to you?" depends on where the person with DS is at the moment of asking. (e.g. when at home: parents, when at work: colleagues) - Scepticism about friendships of their children with DS. - Parent often arranged social activities/meetings for their children with DS.	- Decease of someone near may have large impact on person with DS. - Support staff is only temporarily important for people with DS, they come and go.	- People with DS have a large social network, often arranged by parents. Also loneliness is present. - Decease of someone near may have large impact on person with DS.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Impact DS on others		- "A child with DS teaches you to live in the moment." - "A child with DS costs a lot of time and energy" Health care professional do not always realise that. - Siblings of people with DS often receive less attention than their brother/sister with DS. - If their children with DS live in a living facility, parents are still charged with many tasks (e.g. cleaning their children's apartment). - Many (not all) parents are worried about who will look after their children with DS if they cannot do it anymore or when children leave home.	- It's hard for parents to 'let go' of their children with DS. Hence the worries. Support staff will never be able to look after a person with DS as his/her parents do/ did. - If people with DS become older and function decline occurs, this may impact other residents (e.g. because of changed behaviour)	- Worries of parents (about who will take care about their child with DS) are a real burden to them. - Health care professionals should acknowledge the challenges of having a child with DS.
Patient journey	- Various aids are being used, often mentioned: glasses, arch support. Not always accepted. - Various medications - Various health care professionals involved	- Idem - Care also includes: support, educational services, transportation, social services, and (absence of) financial coverage. - Complex system of services. - Low level of medical knowledge among support staff. - Some examples of complementary / alternative therapy.	- Idem. Additionally, volunteers are mentioned. - Need for more medical knowledge among support staff.	- Aids not always accepted by people with DS, this does not seem to be effective care. - Parents are looking for help in complementary / alternative medicine. - Medical care is one of many other services involved. - Parents are well informed about needed and/or available care, they have become DS-experts.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Quality of care: Person-centred care: general	"If you know a health care professional for some time, you can trust that person, which makes talking easier"	- Trust relationship with health care professional is important. - Waiting (waiting room) may be difficult for people with DS (e.g. because they become nervous). - Health care professionals have to 'click' with a person with DS. - Health care professional who knows about all health problems and can give advice about all conditions together is needed.	A known health care professional is important	- Trust relationship and knowing each other is important: need for continuity of care. - "Clicking" of health care professionals and person with DS is important. - Health care professional should be able to combine information about different conditions and give advice accordingly.
Quality of care: Person-centred care: emotional support / alleviation of fear	Health care professional has to put me at ease, make jokes and ask 'other' (non-medical) questions.	Health care professional has to put person with DS at ease.	Example of putting someone at ease: show what will happen by making support staff undergo a treatment first.	Putting a person with DS at ease is very important.
Quality of care: Person-centred care: communication / information	If a health care professional talks slowly, I can understand him/her better.	- Health care professionals have to: use pictures, repeat questions, provide time for processing text, and take time to listen. - Parent often functions as interpreter for their child with DS.	- Health care professionals have to: make sure a patient feels being seen and heard, make contact (not communicating *about*, but *with* a person with DS). - Support staff often functions as interpreter for person with DS.	- Feeling of being seen and heard is important. - Health care professional should adapt communication (talking pace, time to listen/process) - An 'interpreter' is often needed
Quality of care: Person-centred care: involvement of family/ friends		- Health care professional taking role of "sparring partner" is nice. - Health care professional may confirm you are doing well as a parent.	Parents and support staff do not always agree about what is best for the client with DS. You have to cooperate.	Cooperation between parents, health care professionals and support staff is needed.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Quality of care: Person-centred care: physical comfort	- Fear for being in pain. - Being brought under narcosis for small surgeries	Benefits of a treatment may be different in people with DS as compared to people without DS.	- Alleviation of pain and comfort is especially important in end-of-life care	Careful consideration of burden and benefits of a treatment is needed.
Quality of care: Person-centred care: preferences / values of patient	Often mentioned idea: "You die in hospital"	- Knowing a patient with DS and his/her comorbidities is important. - Health care professional should consider pressure experienced by parents related to managing care. - Parents prefer a holistic approach.	- Health care professionals should connect with internal world of person with DS. - People with DS should be able to make their own choices. - In end-of-life care, needs should be appreciated in every stage.	- Large pressure experienced by parents related to managing care and 'total package'. - Health care professionals should try to align their actions with internal world of people with DS.
Quality of care: Effectiveness	The doctor has to cure me	- Health care professional doesn't always have DS-specific knowledge, but doesn't always need to have it. As long as (s)he really 'sees' the person with DS. - Many health care professionals are able to communicate well. - Some nurses are not able to take blood in people with DS. - Regular health screening (provided by Downteams) is very important - We do not need regular screening, we'll go to a doctor if needed. - Early intervention ® often used for development of children with DS. - Support staff doesn't have time/ knowledge to care for my son/ daughter well	- Little knowledge about DS-specific (health) problems. It is important though. - Support staff plays important role in signalling symptoms and treatment compliance. - Regular screening by Downteams is too much of a snapshot-view about a person with DS. - Behavioural expertise (e.g. psychological care) is very important in care for people with DS.	- No agreement among parents about whether DS-specific expertise is needed - Health care professionals should have better communication skills. - Health screening and/or Downteam is very useful versus it is not needed / takes too much effort. - Behavioural expertise deserves more attention.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Quality of care: Efficiency	"I threw my insoles away"	- Not all care is effective/needed - Not all care providers (in Downteam) provide added value. - Some care is too protocoled. - People with DS often have problems with aids (glasses, insoles)	- Cooperation between service provider and health care professionals not always good. - Hospitals are often not flexible enough to provide good care to people with DS. - People with DS often have problems with aids (glasses, insoles)	- Health care is sometimes too protocoled / inflexible. - In providing medical aids, a more thorough evaluation of person's situation / acceptation seems needed.
Quality of care: Equity		- Good care / support dependent on financial situation / managing skills of parents. - Some health care providers refuse to treat people with DS. - Special treatment is not always needed (people with DS are also just people)	- Some health care professionals don't even try to introduce themselves to people with DS (especially in people with DS with severe intellectual disability) - People with DS easier get needed care than people with intellectual disability (because DS is visible and more commonly known) - Support staff important in advocating for rights of people with DS	- Access to health care not always provided to people with DS, may be dependent of skills of parents/ support staff. - Special treatment not always needed.
Quality of care: Safety	Most people with DS are not in control of medication intake.	Parents question whether support staff notices health problems of their child with DS (in time).	- Hospital staff is sometimes not aware of disabilities of people with DS. - Support staff is often in charge of medication management.	- Medication management needs attention. - Worries of parents regarding expertise of support staff.

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Dealing with complexity of care system	- Transportation for going to hobby or preferred day activity/ work is not always available. - Most people with DS were satisfied with where they lived.	- Medical care is not the problem, it's the rest: handling (changing) regulations, aligning all care and support, (financially) arranging, advocating for needed services, related paperwork, dividing tasks with support staff. - A 'case manager' is lacked. - Volunteers are needed to accompany people with DS to activities. - Good transition from paediatric to adult care is important. - Different home may be needed when people with DS get old. - Many changes in support staff.	- Medics or authorities use (only) IQ to decide about level of needed care. Total functioning is not taken into account. - In most cases, parents (not support staff) accompany clients with DS when visiting health care. - Different idea (compared to parents) about what's best for client with DS - Different home may be needed when people with DS get old. - Many changes in support staff. - Support staff important in informing all people involved about health/treatment of client with DS.	- Managing all needed services is large burden for parents. - Role of case manager should be fulfilled. - Parents and support staff not always agree about what's best for person with DS. - Transitions may be difficult: paediatric → adult, or when getting old.
Health literacy and life style	- Monitoring weight and dieting is often mentioned. - Well informed about health(care) - Several mentioned to be able to cope with pain/burden	- Many parents monitor weight of their (adult) children with DS. - Parents are alert signalling symptoms and are sometimes afraid that support staff is not (enough) alert. - Parents gathered a lot of information about DS (and related health problems)	- Weight and dieting is often mentioned. - Sexuality issues were mentioned.	- Managing weight is important and often a problem. - Parents and people with DS are well informed about DS and related conditions. - Especially parents are alert signalling symptoms and worry whether support staff is alert too. (related to worries under "safety")

Theme	Said by:			Interpretation by authors
	People with DS	*Parents*	*Support staff*	
Information lack, finding, and exchange.		- Would have liked more information on impact of DS on family, educational support, and school choice. - It's often difficult to find right health care provider. A list with available professionals would be helpful.	- Support staff often attends visits to health care in order to make sure all needed information is transferred to the right persons. - Parents attending health care visits do not always share information about their child's condition because they do not want to see deterioration of their child.	- Better information about available care and services would be nice. Parents struggle in finding it. - Support staff important in objective information exchange.
How quality of care may influence quality of life	- Sleep apnoea has been treated, less tired now - Some think it's ok to have glasses/hearing aids/insoles (arch support), others refuse to wear them. - "I go to the physiotherapist every week". - Losing weight is important. - If I'm ill I cannot go to work or hobby. - "my father died in hospital".	- Good health is a prerequisite for quality of life - Aids not always accepted - In some cases, not treating a condition may be better for a person with DS. - Health care / screening may discover physical cause for behavioural problems. - Regular health screening may lead to timely detection of health problems. - People with DS often hide their illness/ burden/ pain, because they do not want to skip work/ hobby. - Health care is sometimes part of weekly structure of person with DS, and therefore important. - Also psychological care may be very helpful. - Good health care should 'see' the individual with DS.	- Good health is a prerequisite for quality of life - Aids not always accepted - In some cases, not treating a condition may be better for a person with DS. - People with DS often hide their illness/ burden/ pain. - If people with DS are treated respectfully, this contributes to their feeling of being 'seen and heard'.	- Good health is a prerequisite for quality of life - Finding the right balance between burden of a treatment and outcome for patients with DS, is important. The balance may be different as compared to general population. - Importance of regular screening: people with DS often hide their illness/ burden/ pain, to explain problematic behaviour. - Illness/ burden/ pain often hidden. - Health care may be part of weekly structure and therefore contributing to well-being. - Psychological care should not be forgotten. - Individual approach should be applied. - Be aware of ideas of people with DS about health care (e.g. "you die in hospital"). - Respectful health care contributes to (mental) well-being of people with DS.


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Capturing the complexity of health care for people with Down syndrome in quality indicators - a Delphi study involving health care professionals and patient organisations

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ABSTRACT

Background: Insight into quality of health care for people with Down Syndrome (DS) is limited. Quality indicators (QIs) can provide this insight. This study aims to find consensus among participants regarding QIs for health care for people with DS.

Methods: We conducted a four-round Delphi study, in which 33 health care professionals involved in health care for people with DS and two patient organisations' representatives in the Netherlands participated. Median and 75-percentiles were used to determine consensus among the answers on 5-point Likert-scales. In each round, participants received an overview of participants' answers from the previous round.

Results: Participants agreed (consensus was achieved) that a QI-set should provide insight into available health care, enable health care improvements, and cover a large diversity of quality domains and health care disciplines. However, the number of QIs in the set should be limited in order to prevent registration burden. Participants were concerned that QIs would make quality information about individual health care professionals publicly available, which would induce judgement of health care professionals and harm quality, instead of improving it.

Conclusions: We unravelled the complexity of capturing health care for people with DS in a QI-set. Patients' rights to relevant information have to be carefully balanced against providers' entitlement to a safe environment in which they can learn and improve. A QI-set should be tailored to different health care disciplines and information systems, and measurement instruments should be suitable for collecting information from people with DS. Results from this study and two preceding studies, will form the basis for the further development of a QI-set.

BACKGROUND

Down syndrome (DS) is the most prevalent genetic cause of intellectual disability (ID) (De Graaf et al., 2017; Phelps et al., 2012). People with DS suffer from a large variety of health problems and therefore have complex health care needs, with many different health care providers involved (Coppus et al., 2017; Grieco et al., 2015; Phelps et al., 2012; Weijerman & De Winter, 2010).

It is widely acknowledged that health care for people with DS should be of high quality in order to meet their specific health care needs (Grieco et al., 2015; Kinnear et al., 2018; Skotko et al., 2013). This is supported by the Convention on the Rights of Persons with Disabilities, advocating high-quality health care for people with disabilities, and acknowledging the right for obtaining the highest possible level of health (UN, 2006). However, little is known about the quality of DS-specialised health care (Lavigne et al., 2015; Van den Driessen Mareeuw et al., 2017).

Quality in health care is multidimensional. The World Health Organization formulated six dimensions of health care quality: 1) effective (evidence-based and based on needs), 2) efficient (maximising resources, avoiding waste), 3) accessible (timely, geographically reasonable, in a suitable setting), 4) acceptable/patient-centred (taking into account preferences, culture of patient), 5) equitable (same level of quality for everyone) and 6) safe (minimising risk and harm) (WHO, 2006).

Quality indicators (QIs) - also known as quality measures (Boulkedid et al., 2011) - are an important tool in health care quality, as they can improve clinical decisions, guide organisational reform, and structure the development of multidisciplinary teams (Donabedian, 2005). Moreover, QIs can provide patients with information that enables them to choose the best suitable care (Delnoij et al., 2010). However, an authors' former study revealed that, up to now, QIs measuring quality of health care for people with DS, do not appear to exist (Lavigne et al., 2015; Van den Driessen Mareeuw et al., 2017). The study found that existing QIs concern people with ID in general (not people with DS in particular), or focus, for instance, on care in assisted living facilities (not specifically on health care) (Lavigne et al., 2015; Van den Driessen Mareeuw et al., 2017).
According to Donabedian's (2005) well-known framework for quality in health care, a QI-set may include different types of QIs: structure, process, and outcome QIs

(Donabedian, 2005; Rademakers et al., 2011). Structure refers to the setting in which health care is provided (e.g. administrative structure), process to how health care is provided (e.g. followed procedures), and outcome to the result of health care provided (e.g. recovery, survival) (Donabedian, 2005). Generally, QIs are based on quality standards, such as guidelines or protocols (Kötter et al., 2012; Mainz, 2003). In the Netherlands, a guideline for multidisciplinary health care for children with DS (Borstlap et al., 2011) is present and is currently being revised. Until now, such a guideline concerning adults with DS has not been present, but is currently being developed.

The present study aims to find consensus among health care professionals and patient organisation representatives regarding QIs for health care for people with DS in the Netherlands. This health care involves, amongst others: a paediatrician, ID physician (in the Netherlands, there is an ID-specialised training for physicians), general practitioner (GP), physiotherapist, speech therapist, psychiatrist, cardiologist, ophthalmologist, and DS-specialised multidisciplinary outpatient clinics, so-called 'Downteams' (Bull, 2011; Coppus et al., 2017; Skotko et al., 2013; Tenenbaum et al., 2008; Weijerman & De Winter, 2010). There are paediatric and adult 'Downteams' in the Netherlands. Paediatric 'Downteams' typically include a visit to the paediatrician, physiotherapist, ENT (ear-nose-throat)-specialist and others, all on the same day. Adult 'Downteams' are still scarce and have a slightly different composition, due to different needs in adulthood.

The present study is part of a larger project aiming to develop a QI-set for health care for people with DS. The project includes a literature review on existing QIs for health care for people with DS (indicating the absence of QIs that could serve as a basis for our QI-set) (Van den Driessen Mareeuw et al., 2017), a qualitative exploration of how people with DS, parents and support staff define quality in health care (Van den Driessen Mareeuw et al., 2020b) (see Table 4.1), and the current study. In the final project step, findings of the three studies will be combined in order to formulate QIs. In the present study, the following research questions are addressed:

1. ***According to health care professionals and patient organisations' representatives, how should a QI-set measuring quality in health care for people with DS be defined?***
   a. *Which purposes should it serve?*

b. Which health care disciplines, services and quality domains should it cover?

c. Which type of QIs (structure, process, outcome) should it include and how many?

2. ***According to health care professionals and patient organisations' representatives, what factors should be taken into account in the further development and implementation of the QI-set?***

Table 4.1 Summary of outcomes of previous study

Outcomes from previous study[a]
Method and participants: Qualitative design including semi-structured interviews with people with DS and with parents, and focus groups with support staff members (of people with DS living in assisted living facilities) *Summary of findings:* - Participants mentioned a large variety of health care and other services people with DS used. Among others: 'Downteam', GP, dentist, psychologist, physiotherapist, speech therapist, ear-nose-throat physician, ophthalmologist, family support, educational support. - According to participants, good health care is: o Person-centred: The person with DS and his/her values and preferences are central; The personal situation and life stage of the person with DS are taken into account and caregivers are involved; Communication between professional and person with DS (and his/her caregivers) is respectful and adapted to the abilities of the person with DS. o Effective, efficient and accessible: Timely recognition of health problems, Health care professionals with DS-expertise are nearby; Information about available care is present. o Multidisciplinary, well-coordinated and integrated: It includes actors outside health care (e.g. school, work); Information is shared (between professionals); Consultations are planned in a synchronized manner; Transition from paediatric to adult health care and services proceeds smoothly.

Abbreviations: DS = Down syndrome; GP = General practitioner.

[a] Qualitative exploration of opinions and experiences of people with DS, parents, and support staff regarding health care quality (Van den Driessen Mareeuw et al., 2020b).

METHODS

A Delphi technique was used in order to achieve consensus among experts in health care for people with DS about relevant items for QIs and related practical issues. Our study is an exploratory inquiry concerning personal opinions of professionals on health care quality. According to Dutch legislation (Wet medisch-wetenschappelijk onderzoek met mensen, article 1, part 1.b.), ethics approval was deemed unnecessary, since participants in our study were not subject to procedures and were not required to follow rules of behaviour. We obtained a written informed consent statement from all participants prior to the study. This allowed us to use participants' contact details for sending them the questionnaires, or for contacting them in case of problems with receiving or filling

out the questionnaires. In this statement, participants also approved the use of their answers to the Delphi-questionnaires in an anonymous manner for the aims of the study.

Participants

We included representatives of all relevant disciplines involved in health care for people with DS and patient organisation representatives, all having expertise in health care for people with DS. This composition is similar to the composition of the working group developing guidelines for health care for people with DS (Borstlap et al., 2011). Recruitment of participants was done by contacting professional organisations from relevant disciplines and two patient organisations (one specific DS organisation and the umbrella organisation of Dutch patient organisations). We explained the purpose of our research and the expected time investment, and asked the organisations to identify members of their organisations with expertise in health care for people with DS. When identified members had agreed to participate, contact details were provided to the researchers, who in turn contacted the members. As the Dutch professional organisation of GPs declined to identify eligible GPs because of other priorities, GPs were recruited via the network of the authors and participants, and/or by using publicly available contact details. Table 4.2 provides an overview of the participant characteristics.

Four-round Delphi procedure

A Delphi study uses a series of questionnaire-rounds in order to establish consensus among a group of experts about a certain topic (Boulkedid et al., 2011; Hsu & Sandford, 2007; Keeney et al., 2006), and is suitable for the selection of QIs (Diamond et al., 2014). In such an iterative process, each next round is based on the participants' answers in the previous round. Only items for which no consensus among participants is found, are presented in the next round. Furthermore, participants receive an overview of the overall group response of the previous round, based on which they can reconsider their initial answers (Diamond et al., 2014; Keeney et al., 2006). Our study consisted of four consecutive rounds:

- **Round 1**: Introduction to themes, initial inventory of level of consensus;
- **Round 2**: Feedback on Round 1 and revisiting themes on which no consensus existed;
- **Round 3**: Exploration of consensus on sub-domains;
- **Round 4:** Final consensus building

We used online questionnaires, which were composed using QualtricsXM®. Online questionnaires allow participants to fill out the questionnaires wherever they want, allow anonymous participation of experts across various locations, and prevent one (or a few) expert(s) from dominating the consensus process (Boulkedid et al., 2011; Hsu & Sandford, 2007).

Table 4.2 Participant characteristics

Characteristic	n=35
Age (y) [mean (stdev) [range]]	50.5 (9.6) [30-73]
Gender [number, (%)]	
Female	32 (91.4%)
Male	3 (9.0%)
Profession	
Audiologist	1 (2.9%)
Dentist (ID-specialised)	3 (8.6%)
Dermatologist	1 (2.9%)
Dietician (ID-specialised)	2 (5.7%)
General Practitioner	2 (5.7%)
ID physician	3 (8.6%)
Municipal Health Services doctor	1 (2.9%)
Nurse / coordinating nurse (ID-specialised)	3 (8.6%)
Occupational therapist	2 (5.7%)
Ophthalmologist	1 (2.9%)
Orthoptist	2 (5.7%)
Paediatrician	2 (5.7%)
(child) Physiotherapist	4 (11.4%)
Psychiatrist (child/youth/adult)	1 (2.9%)
Psychologist	1 (2.9%)
Podiatrist	2 (5.7%)
(child) Rehabilitation physician	1 (2.9%)
Representative of patient organisation	2 (5.7%)
Speech therapist	1 (2.9%)
Time working in this profession (y)	
[mean (stdev) [range]]	19.2 (10,2) [0.7-40]
Frequency of contact with people with DS [number; (%)]	
(almost) daily	9 (25.7%)
Weekly	14 (40.0%)
Monthly	7 (20.0%)
Half-yearly	3 (8.6%)
Yearly	1 (2.9%)
Less than once a year	1 (2.9%)

Abbreviations: y=year(s); stdev=standard deviation; ID=Intellectual Disability

Questionnaires and consensus

All questionnaires contained questions with a five point Likert-scale, multiple choice questions and open-ended questions. Using the Likert-scale questions, participants rated items in terms of relevance for the QI-set (1 'very important', 2 'important', 3 'neutral', 4 'not that important', 5 'not important at all'), or indicated to

what extent they agreed with propositions (1 'totally agree', 2 'agree', 3 'neutral', 4 'disagree', 5 'totally disagree'). In round 1 an 'I don't know'-option was also included. Consensus was defined in advance, as follows: if at least 75% of the participants rated an item as 1 or 2 and the median was ≤ 2, consensus was achieved among the participants about including the item in the QI-set, or about agreeing with a proposition. If 75% of the participants rated an item 4 or 5 and the median was ≥ 4, consensus was achieved among the participants about excluding the item from the QI-set, or about disagreeing with a proposition. In all other situations, it was concluded that consensus was not achieved among participants. Although there is no standard for defining consensus in Delphi studies, using a combination of percentages and median for defining consensus is generally accepted (Boulkedid et al., 2011; Diamond et al., 2014). A 75% cut-off is considered adequate in Delphi studies (Keeney et al., 2006). We decided to present some items to the participants despite the fact that consensus was obtained for these items in the previous round(s), because some participants had not been able to join the first round, or because we thought the items should be presented as a complete set (e.g. all health care disciplines possibly involved in health care for people with DS). If we deemed more detailed information was needed, more specialised items/ propositions, or differently formulated propositions were presented to the participants (e.g. quality domains were presented in round 1 and sub-domains in round 3). The multiple choice questions and the open ended questions allowed participants to explain their 'rated' answers or add relevant QI-items.

The topics of the questionnaires were largely based on outcomes of the previous study investigating the experiences and opinions of people with DS, parents and support staff regarding quality in health care (Van den Driessen Mareeuw et al., 2020b) (see Table 4.1) and on the multidisciplinary guideline for health care for children with DS (Borstlap et al., 2011). Additionally, the questionnaires contained topics related to the development, implementation and use of QIs, informed by literature and expertise of the authors. Topics addressed in the questionnaires and number and type of questions are shown in Table 4.3. An English translation of the questionnaires can be found in Appendix 4-I.

Table 4.3 Topics addressed and type of questions per round

	Topic addressed in:			
Topic addressed	**Round 1** Introduction to themes, initial inventory of level of consensus	**Round 2** Feedback on Round 1 and revisiting themes on which no consensus existed	**Round 3** Exploration of consensus on sub-domains	**Round 4** Final consensus building
Participant characteristics	6 open ended questions (such as age, gender, frequency of contact with people with DS).	Idem: same questions were presented to participants who had not participated in round 1.		
Purpose of QI-set (e.g. transparency, quality improvement, auditing, insurance)	9 purposes, rate importance	12 propositions[a]	9 propositions[a]	
Quality domains to be included in QI-set (e.g. coordinated care, person-centeredness, clinical outcome)	10 items[b] and 1 proposition for children with DS; 10 items[b] and 1 proposition for adults with DS	7 items[b] for children and adults with DS	28 items[b] (sub-domains)	1 proposition[a]
Health care disciplines to be included in QI-set (e.g. Downteam, psychological care, physiotherapy)	14 items[b] and 1 close-ended question for children with DS; 14 items[b] and 1 close-ended question for adults with DS	6 propositions; 30 items[b] for children; 30 items[b] for adults with DS	4 open-ended questions	1 proposition[a]
Number and type (structure / process / outcome) of QIs		2 close-ended questions	2 propositions; 1 close-ended question	2 propositions; 3 open-ended questions
Information sources and transparency of QIs and practical issues regarding development	1 close-ended question; 1 open-ended question	1 proposition; 1 close-ended question; 6 open-ended questions	6 propositions; 1 close-ended question; 2 open-ended question	17 propositions
Health care quality for people with DS and current use of QIs	3 close-ended questions; 3 open-ended questions	15 propositions		
Aim of the study	1 open-ended question			

Abbreviations: DS = Down syndrome; QI = Quality indicator.
Empty fields indicate that the topic was not presented to the participants in the concerning round.
[a] Participants indicated to what extent they agreed with propositions (1 'totally agree', 2 'agree', 3 'neutral', 4 'disagree', 5 'totally disagree').
[b] Participants rated items (i.e. health care disciplines/services or quality domains) indicating the relevance for the QI-set (1 'very important', 2 'important', 3 'neutral', 4 'not that important', 5 'not important at all').

Delphi in one day

The first questionnaire was sent out on April 25th 2018, the other three on May 30th 2018. This timeframe was chosen because participants preferred to conduct the study (predominantly) on one day. This short study duration would thus prevent participant drop-out related to large time intervals between the rounds. It would also limit time investment of both participants and researchers, as participants do not need re-introduction into the topic at the start of each new round, and data collection proceeds quickly. Although the time intervals between the rounds in our study were much shorter than in classic Delphi studies (Keeney et al., 2006), literature does not provide any reason to assume that a shorter study duration affects the results (Blume et al., 2016). However, in order to allow for such short time intervals, the rounds required thorough preparation, enabling participants to fill out the questionnaires swiftly, and enabling researchers to perform analyses and adapt the questionnaires accordingly. Therefore, the authors composed most questions beforehand, by anticipating the possible responses of the participants and by using preliminary insights resulting from round 1. Because of this, only a few questions needed to be newly composed between round 2, 3 and 4, and most questions only had to be moved, slightly rephrased, or removed. Additionally, used software was set ready to quickly provide the researchers with information needed to assess consensus (median and 75-percentiles) and with an overview of open-ended question answers. Furthermore, roles of the research team (i.e. obtaining medians and 75-percentiles; extracting open-ended question answers, chairing the discussions (see next paragraph "Analysis"), adapting and sending out the questionnaires) were allocated beforehand.

Analysis

During the study, we used percentages provided by QualtricsXM® and the median calculated using IBM SPSS Statistics 24, to determine whether the answers of the participants on the Likert-scale questions had resulted in consensus. From the multiple choice questions, only frequencies (percentages) were calculated. Analysis of the answers from open-ended questions included reading and discussing the answers by all authors, which resulted in identification and structuring of key issues. All authors were involved in all iterations of the study, in an e-mail conversation (first round) and in a face-to-face meeting (rounds 2-4).

Afterwards, in order to structure the data, a dataset containing data from all rounds was created using IBM SPSS Statistics 24, and median and 75-percentile of the Likert-scale questions were calculated again. The calculations were done with and without the patient organisation representatives' answers, in order to discover whether their answers differed from the health care professionals' answers. Differences were indicated together with the concerning findings, in order to interpret the results.

RESULTS

Participants flow

A total of 35 eligible participants was identified. However, one participant could not allocate time for participating in any of the rounds and answered only one question in round two and three. Ten participants could not participate in all rounds. Figure 4.1 shows a flowchart of the number of participants per round. On average, participants needed 55, 52, 25 and 14 minutes to complete questionnaires 1, 2, 3 and 4 respectively, with a maximum of 114, 85, 45, and 48 minutes.

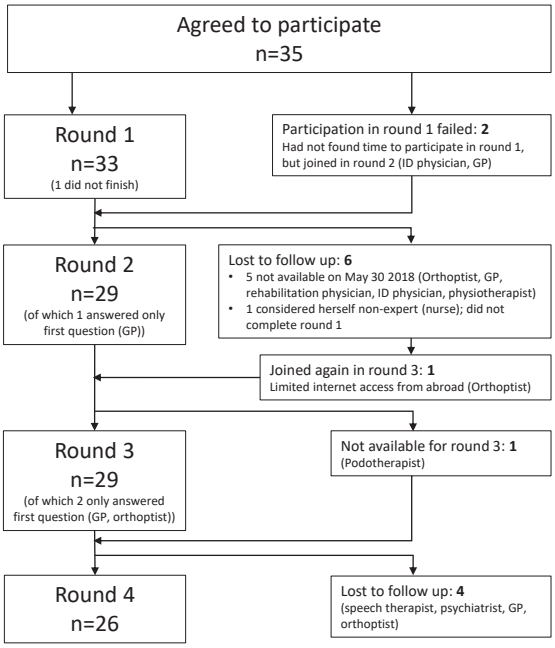

Figure 4.1 Flowchart of number of participants for each Delphi round.

Results Delphi rounds

Distributed across the four rounds, 259 questions were presented to the participants, comprising 20 open-ended questions, 11 closed-ended questions and 228 propositions or items, of which 107 had resulted in consensus among the participants. See Table 4.4.

Table 4.4 Number and types of questions per round and consensus among participants on propositions and items

Round	Total number of questions	Open-ended questions	Closed-ended questions	Propositions / Items	Consensus
Round 1	72	5	6	61	37
Round 2	110	6	3	101	31
Round 3	54	6	2	46	28
Round 4	23	3	0	20	11

Below, the results of the four Delphi rounds are presented in two parts: 1) Defining purposes and identifying QI-topics; and 2) Considerations for further development and implementation of the QI-set. More details about the results can be found in Appendices 4-II, III, IV, V, and VI.

1) Defining purposes and identifying QI-topics

Purposes

In the first three rounds, participants indicated the purpose(s) to be served by the QI-set. See Table 2.5, first row ('Purpose of QIs').

Related to the purpose "provide health care professionals with information on where to find suitable health care (providers)", participants explained that providers could use this information for making referrals. Especially for generalists (such as GPs), who cannot reasonably be expected to have much DS-specialised expertise, QIs could be helpful in identifying specialised health care professionals to refer to.

Additional to the purposes "improving health care on the national level" and "improve health care for people with DS delivered by their organisation (e.g. health centre, hospital, department)", participants mentioned that QIs could be part of audits, and could be used to improve processes (logistics, management, ICT etc.). Furthermore, participants explained that QIs should enable benchmarking of one's own functioning as compared to that of colleagues at individual, regional or national level.

About the purpose "using QIs as input for developing guidelines", consensus was achieved in the first round. However, participants commented that QIs should

not be used as input for guidelines, but rather the other way around (guidelines should define indicators). We therefore decided to present this purpose to the participants in round two again, which did not result in consensus.
Although there was consensus concerning "QIs should be used to reduce differences in quality of provided health care by different providers", some participants argued that differences should exist between providers, because if differences would not exist, this may imply that differences between centres of expertise and other health care providers - very much needed for health care for people with DS – could not exist.

Quality domains

In the first three rounds, participants indicated per quality (sub-)domain how important they considered it to be covered by the QI-set. Table 4.5, second row ('Quality domains') shows the quality domains that, according to consensus among the participants, should be covered by the QI-set.
Although consensus existed regarding including person-centeredness in the QI-set, this was not reflected in participants' answers regarding sub-domains of person-centeredness, presented to the participants in following rounds. On the one hand, participants explained that QIs should measure whether health care is adapted to the needs of the person with DS, which may also increase effectiveness. On the other hand, no consensus existed about: adapting care to the preferences and desires of the person with DS, self-management, considering experienced burden for parents and other caregivers, and organising multidisciplinary appointments on one day.
Furthermore, participants argued that concepts such as quality of life and daily functioning should not appear in the QI-set, because they are too complex to be measured by QIs, too little related to quality of delivered care, or more suitable for inclusion in scientific research, than for being part of a QI-set. Others argued that such concepts should appear in the QI-set, because this would result in increased awareness among health care professionals about these important concepts.

Health care disciplines/services

In round one and two, participants indicated how important they considered each health care discipline or service to appear in the QI-set (see Table 4.5, third row ('Health care disciplines / services')). Participants unanimously indicated that the set should contain one or more QIs on Downteams for children. It was even

argued that a QI for Downteams could function as an indicator for the quality of all other health care for a child with DS, because Downteams are expected to have an overview over the total package of care. However, it was also noted that not all children with DS visit Downteams, implying that a 'Downteam QI' would not be able to indicate quality of health care for all children with DS. A QI measuring quality of care provided by a paediatrician would therefore be more important. Similarly, a QI measuring health care quality of adult Downteams, would not be representative for all health care for adults with DS, since the number of adult Downteams is (too) small, as is the number of ID physicians. Participants explained that GPs sometimes provide the health care that is not provided by ID physicians / adult Downteams. Therefore, including a QI on health care provided by GPs could be important for adults with DS. However, a reason mentioned for *not* including GP-care in the QI-set is that GPs were not expected to have DS-expertise, because they have only a small number of patients with DS.

Furthermore, participants did not agree about coverage of visual functioning and dental care. Monitoring visual functioning was mentioned as a candidate indicator, because visual functioning is apt to change over time. However, no consensus was achieved on including visual screening in the set. Participants' comments about dentistry indicated that some sort of dentistry should be in the QI-set. However, it remains unclear which form of dentistry should be in the QI-set, as some people with DS need a specialised dentist, while for others a general dentist suffices. A mentioned reason for including a QI measuring specialised dental care, was based on the idea that a specialised dentist should always be involved, in order to monitor, recognise and treat DS-specific dental problems.

There was a lot of discussion about including non-medical disciplines/services in the QI-set. For example, consensus about including 'family support' was only achieved when the patient organisations' representatives were included in the analysis, and there was no consensus about including support staff of assisted living facilities in the QI-set. Moreover, the proposition "QIs should also cover non-medical disciplines" did not result in consensus. Some participants argued that including them was especially important because it is too much of a blind spot among health care professionals, whereas others explained that non-medical disciplines/services do not belong to a QI-set for quality of health care.

Table 4.5 Summary of findings: Defining purposes and identifying QI-topics

Theme	Consensus about (Likert-scale questions)	Round(s) in which theme was addressed
Purpose of QIs	QIs should: • provide people with DS and their caregivers with information on where to find suitable health care (providers); • provide health care professionals with information on where to find suitable health care (providers); • be used to improve health care for people with DS on a national level; • be used to improve health care for people with DS delivered by their organisation (e.g. health centre, hospital, department), by using the QIs as input for (interdisciplinary) reflective meetings with colleagues, for short term evaluation of health care delivery on the patient level[a], or for adapting protocols; • be used as input for developing guidelines; • be used for inspection and control by national/governmental or intra-organisational authorities; and • be used to reduce differences in quality of provided health care by different providers	1,2,3 (more detailed information in Appendix 4-II)
Quality domains	The QI-set should cover: • Coordination (both within and between organisations and disciplines) of health care for people with DS, including professional collaboration and agreements, and professional-caregiver collaboration; • Transition from paediatric towards adult health care; • Effectiveness, including expertise of health care professionals and timely detection of health problems; • Person-centeredness, including the social system of a person with DS[a]. • Quality of life, daily functioning, autonomy, and participation in society; • Safety; • Clinical outcomes (e.g. blood screening); and • Adherence to guidelines.	1,2,3 (more detailed information in Appendix 4-III)
Health care disciplines / services	• Concerning children, the QI-set should include: Downteam, paediatrics, physiotherapy, speech therapy, dietetics, psychological/psychiatric care, dental hygiene, specialised dentistry, audiology (screening), and family support[b]; • Concerning adults, the QI-set should include: Downteam, ID physician, dietetics, psychological/ psychiatric care, dental hygiene, dentistry, palliative/geriatric care, general practitioner, audiology, and a case-manager. • QI-set should be sensitive to different health care needs in different life phases	1,2 (more detailed information in Appendix 4-IV)
Number of QIs in set	• QIs should include all disciplines involved in health care for people with DS • The QI-set should contain a basic set and additional specialised modules • Each module should contain a maximum of ten QIs • Disciplines are more important to be included in the QI-set if: o more people with DS need them o they contribute more to QoL o there are more doubts about the quality provided by the discipline	2,3,4 (more detailed information in Appendices 4-IV and 4-V)
Type (structure / process / outcome) of QIs in set	The QI-set should include an (almost) evenly distributed amount of structure, process and outcome QIs.	2,3 (more detailed information in Appendix 4-V)

Abbreviations: DS=Down syndrome; QI=quality indicator; ID=Intellectual disability QoL=Quality of life.

[a] Only consensus if patient organisation representatives were left out of analysis.

[b] No consensus if patient organisation representatives were left out of analysis.

Although participants considered adherence to medical guidelines to be an important QI, they also noted that deviation from guidelines may be necessary in order to provide care that answers to the needs of people with DS. Hence, non-adherence to guidelines does not necessarily indicate low quality.

Number and type of QIs

Table 4.5, fourth row ('Number of QIs in set') shows that participants preferred to include all disciplines/services involved in health care for people with DS in the QI-set. However, participants also noted that this would result in a QI-set with too many QIs, leading to a too high administrative burden for the users of the QI-set. In round two, participants thought that the total number of QIs in the set should be, or should not exceed, ten. In round three, participants agreed (consensus) that the QI-set should consist of modules: a basic module containing QIs relevant for all people with DS, and additional modules for specific patient groups or health care services. In round four, participants thought that each module should contain about ten QIs.

In round two and three, participants indicated that they thought the QI-set should contain structure, process, and outcome QIs (see Table 4.5, fifth row ('Type of QIs in set')). They also argued that the number of outcome indicators should be the highest, followed by process and structure indicators respectively.

2) Considerations for further development and implementation of the QI-set

Current and future use of indicators

In round one, the majority of the participants indicated that they expected their colleagues (from the same profession) to be willing to register (extra) data for the QI-set. See Table 4.6, first row ('Willingness to register'). Participants explained that whether or not health care professionals would register data for this QI-set, would be dependent on available time, awareness about the QIs, considered utility of QIs, and frequency of contact with people with DS.

In round one, we also asked participants what kind of quality information they or their organisation currently collected. See Table 4.6, second row ('Current collection of data by own organisation'). Most participants (41%) indicated that their organisation did not collect any quality information. If information was being collected, it primarily concerned information about adherence to

guidelines, clinical outcomes, and findability of the organisation. Furthermore, most participants indicated that they did not use indicators in their work, and if they did use them, it concerned QIs regarding general (not DS-specific) internal improvement of health care or audits (see Table 4.6 third row ('Current use of QIs')). We also asked participants about the guidelines they currently used in their work (see Table 4.6, fourth row ('Current use of guidelines')). The Dutch multidisciplinary medical guideline for children with DS (Borstlap et al., 2011) was the most often mentioned guideline.

Participants were not always in favour of participating in a QI-set that would make quality information publicly available, especially if a QI-set would reveal quality information on the level of individual health care professionals. In round one, participants explained that such information would possibly result in long waiting lists for 'good' providers or professionals, which may in turn negatively affect quality. Moreover, once a health care provider or professional is labelled as 'not good', this would possibly affect the choice of patients for this provider or professional for a long period of time. Because of these considerations, clarifying propositions were presented to the participants in rounds three and four (see Table 4.6, last row ('Transparency')). This confirmed the reluctance of participants to publish quality information (provided by the QIs) about individual professionals. It also showed that participants preferred access to this individual information to be limited to health care providers, in order to prevent judgement of health care professionals by patients or other parties. It should be used for internal improvements instead. Accordingly, participants explained to be reluctant to introduce a quality mark for health care providers. However, other participants argued that a QI-set would enable health care providers/organisations to profile themselves as 'good' health care providers, by 'signing up' for participating in the QIs, on a voluntary basis. Participation in the QI-set would be an indication of DS-expertise, which would also provide insight into available health care for people with DS to caregivers and health care professionals.

Table 4.6 Summary of findings: current and future use of indicators

Theme	Answers to multiple choice / open questions (first 4 rows) and one Likert-scale question (last row)	Number (%) of participants	Round(s) in which theme was addressed
Willingness to register	• My colleagues (from the same profession) will not be willing to register (extra) data for the QI-set	5[a] (16%)	1 (n=32)
	• My colleagues will only be willing to register (extra) data for the QI-set if this would only mean 'clicking a few extra boxes'	14[b] (44%)	
	• My colleagues will be willing to register (extra) data.	13[c] (41%)	
Current collection of data by own organisation	• Information on adherence to guidelines	10 (31%)	1 (n=32)
	• Transition from paediatric to adult health care	3 (9%)	
	• Clinical outcomes	10 (31%)	
	• Quality of life / daily functioning / participation	9 (28%)	
	• Coordination within the organisation	5 (16%)	
	• Coordination between organisations/ disciplines	1 (0%)	
	• Whether organisation is findable for potential patients	4 (10%)	
	• Accessibility	6 (19%)	
	• Expertise of health care professionals	7 (22%)	
	• Person-centeredness	9 (19%)	
	• Equity	4 (10%)	
	• No quality information collected	13 (41%)	
	• N/A	5 (16%)	
Current use of QIs	• Indicators regarding general internal improvement of health care (non DS-specific) or audits,	11 (34%)	1 (n=32)
	• Indicators regarding client satisfaction,	6 (19%)	
	• Indicators regarding discipline/condition-specific (non DS-specific) issues	4[g] (13%)	
	• No indicators	11 (34%)	
	• N/A	3[h] (9%)	
Current use of guidelines	• The multidisciplinary medical guideline for children with DS	13 (38%)	1 (n=32)
	• A general guideline for adults with DS, developed by the organisation I work for	2 (6%)	
	• Discipline-specific guideline(s) for the general population	7[d] (22%)	
	• Discipline-specific guideline(s) for people with ID	4[e] (13%)	
	• Discipline-specific guideline(s) for people with DS	7[f] (22%)	
	• No guidelines	4 (13%)	
Transparency	• QIs should provide quality information on departmental or organisational level (not on individual professionals' level) • Providers should be obliged to publish this quality information on their websites, if they want to be seen as 'DS-specialised'. • QIs should stimulate health care improvement, not judge health care professionals • Privacy of professionals should be protected just as much as privacy of patients.	Percentages are not applicable: consensus was achieved	3 (n=29), 4 (n=26) (more detailed information in Appendix 4-VI)

Abbreviations: DS=Down syndrome; QI=quality indicator; ID=Intellectual disability.
[a] child physiotherapist, dermatologist, GP, ID physician, psychiatrist
[b] audiologist, 2 podiatrists, ID physician, ID-specialised dentist, municipal health services doctor, 2 occupational therapists, ophthalmologist, 2 orthoptists, paediatrician, rehabilitation specialist, speech therapist
[c] 2 dieticians, 2 ID-specialised dentists, 2 ID-specialised nurses, paediatrician, 3 (child) physiotherapists, psychologist, and the two patient organisation representatives
[d] GP, occupational therapy, dermatology
[e] dentistry, dietetics, dementia
[f] physiotherapy for children, speech therapy for children, municipal health service
[g] dentistry, dermatology, cataract, thyroid
[h] the two patient organisation representatives and one retired participant

Data source and development of QIs

Electronic medical records (EMRs) and patient/parent questionnaires were considered the most important information sources for the QI-set. At the same time, participants underlined that both health care professionals and people with DS and their caregivers should not be overcharged with registration burden. See Table 4.7, first row ('Data source'). Participants suggested to transform (a) patient/parent questionnaire(s) into an easy-to-understand app in order to make it suitable for people with DS. Ideally, such an app should be linked to the information system (EMR) in order to store all information together. However, participants identified the large number of existing information systems, often not mutually communicating, as a potential barrier for implementation of a QI-set.

According to the participants, development of the QIs should be done by researchers (the authors) together with all stakeholders. See Table 4.7, second row ('Development of QIs'). Participants mentioned representatives of the same diversity of disciplines as mentioned under 'health care disciplines/services' to be involved in the development of the QIs. It was also noted that it would be difficult to weigh the different opinions of those involved. The majority of the participants (59%) indicated that whether or not they themselves were willing to participate in development of the QIs depended on the time and effort needed.

Table 4.7 Summary of findings: data source and development of QIs

Theme	Answers to multiple choice / open questions (rows 1 & 3) and one Likert-scale question (row 2)	Number (%) of participants	Round(s) in which theme was addressed
Data source	- Data for the QIs should be extracted from the electronic medical records of patients	26 (81%)	1 (n=32)
	- Data for the QIs should be obtained via questionnaires for patients/parents.	25 (78%)	
	- Burden for people with DS and their caregivers should be as low as possible when measuring quality; - People with DS/caregivers as well as health care professionals should deliver information for the QIs; - Parents/other caregivers should themselves be responsible for documenting and keeping track of needed health care for the person with DS; - When people with DS are not able to provide quality information themselves, their legal representative should decide who is eligible to provide this information. - A dialogue between health care professional and person with DS can be used as instrument for measuring customer satisfaction[a]	Percentages are not applicable: consensus was achieved	4 (n=26) (more detailed information in Appendix 4-VI)
Development of QIs	- With involvement of people with DS	23 (83%)	2 (n=28)
	- With involvement of parents/caregivers	26 (93%)	
	- With involvement of health care professionals	27 (97%)	
	- With involvement of health insurers	6 (21%)	
	- I am willing to participate in development	9 (31%)	
	- Whether I am willing to participate depends on the time and effort needed for participation	17 (59%)	
	- I am not willing to participate	3 (10%)	

Abbreviations: DS=Down syndrome; QI=quality indicator; ID=Intellectual disability.
[a] There was only consensus among the participants about this proposition if the patient representatives were left out of the analysis.

DISCUSSION

In this study we aimed to prefigure quality indicators for health care for people with Down syndrome. We used a Delphi technique involving health care professionals and patient organisations' representatives. The findings of this study, together with findings from two previous studies of the authors (a literature review on existing QIs and a qualitative study involving people with DS and their caregivers (Van den Driessen Mareeuw et al., 2017; Van den Driessen Mareeuw et al., 2020b), will be used to inform the further development and implementation of the QI-set.

According to the participants in the current study, QIs should be suitable to inform health care quality improvement, and should be able to provide an overview of available health care to people with DS and their caregivers, and to health care professionals. Participants stressed that QIs should not be used to judge health care professionals. Furthermore, they opted for an evenly distributed mix of structure, process, and outcome QIs, covering the following quality domains: coordination and continuity of health care, effectiveness, safety, person-centeredness, and outcomes concerning health and quality of life. Additionally, participants argued that the QIs should cover all health care disciplines involved in health care for people with DS. However, they urged to keep the number of QIs low, in order to prevent (administrative) burden for health care professionals and people with DS and/or caregivers. Furthermore, development of QIs should be done with involvement of all relevant stakeholders.

Quality improvement and well-informed choices

According to the participants in our study, two key purposes of a QI-set for health care for people with DS are 1) to improve quality in health care and 2) to increase insight into available health care, enabling people with DS (and their caregivers) to make well-informed health care choices, and supporting health care professionals to make well-informed referrals. However, participants in the current study argue that the two purposes may conflict with each other. They explained that if quality information was publicly available, especially when it concerned information on the level of individual providers, a "shaming-and-blaming" situation would emerge. They were concerned that this would hamper quality of care, instead of improve it. A study addressing Parkinson's disease, showed a similar reticent attitude amongst health care professionals towards sharing quality information with patients (Damman et al., 2019). On the other hand, current movements in practice and literature have shown the need for encouraging patients to make well-informed health care choices, although the influence of QIs on health care choices made by patients has been shown to be limited (Damman et al., 2019; Victoor et al., 2016; Zwijnenberg et al., 2016). Hence, patients' rights to relevant information, fostering the choice for the best suitable health care, have to be carefully balanced against providers' entitlement to a safe environment in which they can learn and improve.

Capturing complexity

There was much discussion about defining the coverage of the QI-set. Some participants preferred to include only medical QIs, whereas others were convinced that a QI-set should cover disciplines/services outside health care, such as support staff of assisted living facilities, in order to reflect the complexity of health care for people with DS (Capone et al., 2018; Weijerman & De Winter, 2010). However, based on our results (achieved consensus) we conclude that participants prefer to limit the coverage of the QI-set to the medical domain (including psychological care). This medical focus may be a reflection of the specialised focus of health care professionals and their training, or of the fragmented care system in the Netherlands (O'Hare et al., 2016; Otte-Trojel et al., 2015). Another explanation for this medical focus may be found in social psychology (Ajzen, 2002; Chen et al., 2016): health care professionals may consider quality improvement or transparency within the medical domain within their control, while they consider other domains beyond their sphere of influence and therefore less important for a QI-set. The medical focus may however also be a result of the participants' reluctance to face a high registration burden, which participants repeatedly expressed during the study. This confirms the general understanding that QI-sets should be concise to foster their actual use (Kelley & Hurst, 2018; Westby et al., 2018).

However, even if the coverage of the QI-set will be limited to the medical domain, it will, due to the multi-morbidity related to DS (Capone et al., 2018; Weijerman & De Winter, 2010), include a lot of different disciplines, and many quality domains. Hence, developing a concise QI-set will be challenging, even more so as not all quality domains may be applicable to all disciplines and contexts, and the QI-set will have to be compatible with a large variety of data registration systems used by the different health care providers involved. In order to limit registration burden, registration of data for a QI-set should be possible together with other currently registered data in the electronic medical record (EMR). This would also prevent registration of the same data in separate registries (De Boer et al., 2018), and facilitate data collection (i.e. extraction from information systems) for the QI-set. Literature shows that automated extraction of indicators from EMRs is possible, however, the structure of information systems and the accuracy of registration by professionals is not always sufficient for enabling automated extraction (Borusiak et al., 2018; Verheij et al., 2018). Nevertheless, most participants in our study thought that their colleagues (of the same profession) would be willing to register extra QI-data, especially if registration efforts would be kept as small as possible.

Patient reported information

Participants also suggested to use patient reported information (for example from questionnaires) as input for the QI-set, which should ideally be stored within the EMR, together with the data registered by health care professionals. Such patient information is often obtained using Patient Reported Outcome Measures (PROMs) and/or Patient Reported Experience Measures (PREMs) (Breckenridge et al., 2015; Manary et al., 2013). PROMs focus on measuring outcomes of treatments related to patient functioning, while PREMs address patient experiences regarding health care processes (Westby et al., 2018; Verheij et al., 2018). PREMs/PROMs are considered robust quality measures (Manary et al., 2013). However, due to their cognitive abilities (Grieco et al., 2015), people with DS may not always be able to provide patient reported information, in which case proxies (such as parents) will have to provide this information (Balboni et al., 2013; Schmidt et al., 2010). Nevertheless, patient involvement in health care is considered increasingly important in delivering high quality health care in general (Doekhie et al., 2018), and concerning people with ID (Flynn et al., 2016). It may therefore be worthwhile to explore other ways to obtain information from people with DS that could be used for quality improvements. Examples are using narratives for evaluation (Abma & Widdershoven, 2005) or apps especially designed for people with DS/ID (Kramer & Schwartz, 2017).

Strengths and limitations

The selection of participants reflected the large variety of health care providers involved in health care for people with DS and included two patient organisations' representatives. Although this presumably led to heterogeneity in answers, which may complicate the formulation of QIs, it can be considered a strength of the study. Participant heterogeneity enriches the results of a Delphi study, which enhances the credibility and acceptance of resulting QIs (Boulkedid et al., 2011).

Another strength of the study is that consensus was defined in advance (Boulkedid et al., 2011; Keeney et al., 2006; Diamond et al., 2014) (median ≤ 2 in combination with a 75% cut-off).

The fact that the members of the research team (i.e. the authors) have been collaborating before, may have led to some advantageous knowledge of each other's ideas, which may have affected the research team's discussions, and in turn, the content validity of the Delphi-questionnaires. However, we expect this

effect to be small because of the heterogeneity of the research team (see "Authors' information") and the limited contact frequency of the team members before the study. Moreover, the fact that consensus was defined in advance, improves reliability of the questionnaire results.

There was variation among the participants regarding the time they had been working in their current position, but they represented ample DS-related experience: 91.4% of the participants had been working in their current position for more than seven years; 85.7% had at least monthly contact with clients with DS. Unfortunately, GPs, playing a key role in health care for people (especially adults) with DS (Bakker-van Gijssel et al., 2017), were underrepresented. Despite extensive attempts, we were only able to include one GP, who could only participate in round one.

The time intervals between the rounds in our study were much smaller than in classic Delphi studies, which have a total study duration of three to twelve months (Keeney et al., 2006). The short time-intervals were chosen after consulting the participants about their preferences for taking part in the study, in order to limit participant drop-out. Nevertheless, we could not prevent a drop-out of about 25%. However, a response rate of about 75% is considered quite high in Delphi-studies (Keeney et al., 2006). This relatively high rate was probably achieved by the personal touch we applied in communication with our participants, which is mentioned to be crucial in limiting drop-out (Keeney et al., 2006). A possible disadvantage of the short time intervals may be that it entails limited time for analysis and preparation of questions for next rounds. We mitigated this possible effect on data collection and results by preparing a large part of the questions for successive rounds in advance. Another possible disadvantage of short time intervals is related to the fact that participants have less time to reflect on, and adapt, their answers. However, we considered the questionnaires suitable to be completed within short time intervals, as the complexity of the questions presented to the participants was quite low. This is supported by the fact that the participants in our study completed the questionnaires within reasonable time. Moreover, the most complex questions, which may require much reflection time, were placed in the first questionnaire, which participants had to complete within several weeks (instead of within several minutes for the other questionnaires).

Conclusions

Our study showed the complexity of capturing health care for people with DS in a QI-set that is relevant for both health care providers and people with DS plus their caregivers. We have taken a solid step in unravelling this complexity and its possible impact on developing QIs, thereby making substantial progress in the development of QIs for health care for people with DS. Future research can (and will) build further on this foundation.

Since our study involves a large variety of health care professionals, with heterogenic viewpoints, our findings may not only be relevant to health care for people with DS, but probably to any health care discipline. It is even argued that, because of the complexity of health care for people with DS, the DS population could be used to assess the quality of the health care system in general (Phelps et al., 2012).

Several important lessons from this study should be taken into account in the further development of a QI-set for health care for people with DS. First, our findings indicate that a QI-set for health care for people with DS has two main purposes: it should be suitable for 1) identifying possibilities for improvement of health care for people with DS; and 2) for supporting patients and providers in choosing appropriate health care (providers). However, the two purposes need to be carefully balanced, as extensive information transparency fostering patients' health care choices, may conflict with ensuring safe and supportive working environments for health care professionals, and with fair comparison of providers. Second, capturing health care for people with DS in a QI-set requires the set to be suitable for use by all different disciplines involved, and to be compatible with different information systems. At the same time, the set has to be as concise and compact as possible, in order to limit administrative burden. Third, measurement instruments providing information for a QI-set should be suitable for collecting information from people with DS and their caregivers.

APPENDICES

Appendix 4-I

English translation of the questionnaires of round 1, 2, 3 and 4.

(Original questionnaires are in Dutch)

Round 1

Dear participant,
Thank you very much for your willingness to take part in this study. This study concerns quality indicators for measuring quality of health care for people with Down syndrome. The aim of this study is to identify potential quality indicators and to reveal how these indicators may be used in practice. The study involves four rounds: the current one and three on May 30. The first round entails the current questionnaire, which you are about to start in a few clicks. Please complete this first questionnaire before May 14.

You gave informed consent for participation in this study. Please note that participation is on a voluntary basis. You are free to withdraw from the study at any moment, without an explicit reason.

Please do not hesitate to contact us if you experience any problems [phone number and e-mail address of first author].
Good luck!

Kind regards,
[names of the authors]

Please click "next" to start the questionnaire.

1. What is your age? (scroll down menu 20-90)

2. Wat is your gender?
   - o Male
   - o Female

3. What is your current professional position?
If you are currently not employed, please mention this in your answer and indicate the position you have had for the longest period.
…

4. (approximately) how long did you work / have you been working in this position?
If you are currently not employed, please indicate how long you worked in the position indicated in the former question.
…

5a. Professionally, how often are you in contact with people with Down syndrome (children and/or adults)?
If you are currently not employed, please indicate how often you were professionally in contact with people with Down syndrome in the position mentioned in the former question.
   - o (almost) daily
   - o Weekly

- o Monthly
- o Biannually
- o Annually
- o Less than once a year

5b. Explanation related to your profession and/or your contact with people with Down syndrome (questions 3-5)
(optional)
…

Indicators
Based on literature, existing guidelines, previous input of health care professionals and interviews with people with Down syndrome, their parents, and support staff, we identified relevant elements of quality of health care for people with Down syndrome. The number of elements appeared to be large.

"An indicator is a 'measurable element of practice performance (…) that can be used to assess the quality, and hence change in quality, of care provided" (Lawrence et al., 1997). An indicator is a signalling agent: it is not a direct measure of quality, but indicates a certain aspect of healthcare provision, which may be reason for further investigation. (Handleiding indicatorenontwikkeling, Kennisinstituut Medisch Specialisten, 2013)

Preferably, a small number of indicators provides as much information as possible. In other words, we strive to obtain a good impression of quality of health care for people with Down syndrome using only a few indicators. An important reason for this is to limit administrative burden. Therefore, this study aims to select indicators that best reflect health care quality, and at the same time, lead to the least administrative burden.
However, this study starts extensively, with a broad variety of topics to be potentially measured by the indicators. With the following questionnaires, we aim to reveal the topics that are, according to you, relevant for a set of indicators for health care for people with Down syndrome. In the following questions, a large variety of topics is presented to you. For each topic, you are asked to indicate how important you think it is (the extent to which you think the topic should be reflected in the set of indicators). You are asked to do this for health care for both children (0-17 years of age) and adults (18 years of age and older). You will be able to explain your answers if desired.

The following questions concern health care for CHILDREN (0-17 years of age) with Down syndrome.

6. Health care for children (0-17 years of age) with Down syndrome:

6a. Which elements of health care should be reflected in the set of indicators?

	Very important	Important	Neutral	Not that important	Not important at all	I don't know	Explain your answer (optional)
Downteam (a coordinated team of collaborating multidisciplinary health care providers for children with Down syndrome (0-17y))	O	O	O	O	O	O	…
Paediatrician	O	O	O	O	O	O	…
Physiotherapy	O	O	O	O	O	O	…
Speech therapist	O	O	O	O	O	O	…
Dietician	O	O	O	O	O	O	…
Occupational therapy	O	O	O	O	O	O	…
Podiatrist	O	O	O	O	O	O	…
Dermatology	O	O	O	O	O	O	…
Mental health care	O	O	O	O	O	O	…
Youth health care (municipal health service)	O	O	O	O	O	O	…
Dental care: Dental hygienist	O	O	O	O	O	O	…
Dental care: regular dentist (primary care)	O	O	O	O	O	O	…
Dental care: paediatric / specialised dentist	O	O	O	O	O	O	…
General practitioner	O	O	O	O	O	O	…

6b. Which additional elements of health care should be reflected in the set of indicators? (in other words, do you miss elements of health care / disciplines in the above list and how important do you consider them to be?

PLEASE NOTE: this question (still) concerns children with Down syndrome. (questions concerning adults will follow)

(If you do not want to add elements of health care / disciplines, please click "next").

Add health care element(s) below	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…

7. Health care for children (0-17 years of age) with Down syndrome:

7a. Do you think the set of indicators should be able to measure adherence to the NVK-guideline? (We mean the guideline of the Dutch Paediatrician society (NVK) of 2011: "Een update van de mulitdisciplinaire richtlijn voor de medische begeleiding van kinderen met Downsyndroom" [An update of the multidisciplinary guideline for the medical support of children with Down syndrome]. This guideline is currently being revised. We assume that your answers also apply to the revised version of the guideline. Please find the guideline and its summary here. [weblink to guideline]

	Very important	Important	Neutral	Not that important	Not important at all	I don't know
The indicator set should be able to measure adherence to the NVK-guideline	O	O	O	O	O	O

[IF answer to 7a. was "I don't know", "not important et al", or "not that important":]

7b. Please explain your answer
…

[IF answer to 7a. was "very important", "important", or "neutral":]

7c. Which elements of the NVK-guideline should be reflected in the indicator set? (more than one answer possible)

- O Visit to a paediatrician (frequency with which a child with down syndrome (and his/her parents) visits a paediatrician)
- O Visit to a Downteam (frequency with which a child with down syndrome (and his/her parents) visits a Downteam)
- O Visit to an ENT-physician (frequency with which a child with down syndrome (and his/her parents) visits an ENT-physician)
- O Visit to an ophthalmologist (frequency with which a child with down syndrome (and his/her parents) visits an ophthalmologist)
- O Visit to an orthoptist (frequency with which a child with down syndrome (and his/her parents) visits an orthoptist)
- O Visit to a dentist (frequency with which a child with down syndrome (and his/her parents) visits a dentist)
- O Visit to an orthodontist (frequency with which a child with down syndrome (and his/her parents) visits an orthodontist)
- O Visit to a physiotherapist (frequency with which a child with down syndrome (and his/her parents) visits a physiotherapist)
- O Visit to a speech therapist (frequency with which a child with down syndrome (and his/her parents) visits a speech therapist)
- O Visit to youth care (frequency with which a child with down syndrome (and his/her parents) visits youth care)
- O Whether a heart echo is made
- O Thyroid screening
- O Coeliac disease screening
- O Other elements / topics, namely: ……
- O I don't know

[IF answer to 7a. was "very important", "important", or "neutral":]

7d. Please explain your answer(s) to question 7c. (optional)
…

8. Health care for children (0-17 years of age) with Down syndrome:

8a. Do you think that the set of indicators should include indicators reflecting the use of additional guidelines, protocols and / or quality standards?

	Very important	Important	Neutral	Not that important	Not important at all	I don't know
The indicator set should be able to measure adherence to additional guidelines / protocols / quality standards	O	O	O	O	O	O

[IF answer to 8a. was "very important", "important", or "neutral":]

8b. Which additional guidelines / protocols / quality standards?
(For example: guidelines in your field of expertise, or from the Dutch quality framework for care for people with intellectual disabilities, or other)
…

8c. Please explain (optional)
…

9. Health care for children (0-17 years of age) with Down syndrome:

9a. Which quality domains or topics should be reflected in the set of indicators?

	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
Clinical outcomes (for example: improved serology, heart function, BMI)	O	O	O	O	O	…
Outcomes relevant for the patient (e.g. quality of life, daily functioning, participation)	O	O	O	O	O	…
Coordination *within* an organisation or department (for example: presence of a coordination, multidisciplinary consultation)	O	O	O	O	O	…

	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
Coordination *between* health care professionals of different organisations and sectors (for example: between health care professionals in primary care, secondary care, or social care)	O	O	O	O	O	…
Transition 18- to 18+ (transition / transfer from paediatric to adult health care)	O	O	O	O	O	…
Findability (information on available health care providers and their differences)	O	O	O	O	O	…
Accessibility (for example: ease of making appointments, waiting time, geographical location)	O	O	O	O	O	…
Expertise (for example knowledge present among health care professionals)	O	O	O	O	O	…
Person-centeredness (for example: patient-professional relation, communication, taking into account life phase and preferences of patient and parents)	O	O	O	O	O	…
Equality	O	O	O	O	O	…

9b. Which additional quality domains or topics should be reflected in the set of indicators? (in other words, do you miss quality domains or topics in the above list and how important do you consider them to be?

PLEASE NOTE: this question (still) concerns <u>children</u> with Down syndrome. (Questions concerning adults will follow)

(If you do not want to add quality domains or topics, please click "next").

Add quality domains or topics below	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…

The following questions concern health care for adults (18 years and older) with Down syndrome.

10. Health care for adults (18 years and older) with Down syndrome:

10a. Which elements of health care or disciplines should be reflected in the set of indicators?

	Very important	Important	Neutral	Not that important	Not important at all	I don't know	Explain your answer (optional)
Downteam (a coordinated team of collaborating multidisciplinary health care professionals for adults with Down syndrome (≥18y))	O	O	O	O	O	O	…
ID (intellectual disability) physician	O	O	O	O	O	O	…
Psychologist	O	O	O	O	O	O	…
Dietician	O	O	O	O	O	O	…
Physiotherapy	O	O	O	O	O	O	…
Speech therapist	O	O	O	O	O	O	…
Occupational therapy	O	O	O	O	O	O	…
Podiatrist	O	O	O	O	O	O	…
Dermatology	O	O	O	O	O	O	…
Mental health care	O	O	O	O	O	O	…
Youth health care (municipal health service)	O	O	O	O	O	O	…
Dental care: Dental hygienist	O	O	O	O	O	O	…
Dental care: regular dentist (primary care)	O	O	O	O	O	O	…
Dental care: paediatric / specialised dentist	O	O	O	O	O	O	…
General practitioner	O	O	O	O	O	O	…
Palliative care	O	O	O	O	O	O	…

10b. Which additional elements of health care or disciplines should be reflected in the set of indicators? (in other words, do you miss elements of health care / disciplines in the above list and how important do you consider them to be?

PLEASE NOTE: this question (still) concerns adults with Down syndrome.

(If you do not want to add elements of health care / disciplines, please click "next").

Add health care element(s) below	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…

11. Health care for adults (18 years and older) with Down syndrome:

11a. Do you think the set of indicators should be able to measure adherence to a multidisciplinary guideline for health care for adults with Down syndrome?

NOTE: A multidisciplinary guideline for health care for adults with Down syndrome is currently being developed (Similar to the guideline for children). This question concerns the future use of this guideline.

	Very important	Important	Neutral	Not that important	Not important at all	I don't know
The indicator set should be able to measure adherence to a multidisciplinary guideline describing health care for adults with Down syndrome.	O	O	O	O	O	O

[IF answer to 11a. was "I don't know", "not important et all", or "not that important":]

11b. Please explain your answer (optional)
…

[IF answer to 11a. was "very important", "important", or "neutral":]

11c. A guideline describing multidisciplinary health care for adults with Down syndrome is currently being developed. Which health care elements should this guideline contain AND should be reflected in the indicator set? (more than one answer possible)

O Visit to an ID physician (frequency with which an adult with down syndrome (and his/her relatives) visits an ID physician)

- O Visit to a Downteam (frequency with which an adult with down syndrome (and his/her relatives) visits a Downteam)
- O Visit to an ENT-physician (frequency with which an adult with down syndrome (and his/her relatives) visits an ENT-physician)
- O Visit to an ophthalmologist (frequency with which an adult with down syndrome (and his/her relatives) visits an ophthalmologist)
- O Visit to an orthoptist (frequency with which an adult with down syndrome (and his/her relatives) visits an orthoptist)
- O Visit to a dentist (frequency with which an adult with down syndrome (and his/her relatives) visits a dentist)
- O Visit to an orthodontist (frequency with which an adult with down syndrome (and his/her relatives) visits an orthodontist)
- O Visit to a physiotherapist (frequency with which an adult with down syndrome (and his/her relatives) visits a physiotherapist)
- O Visit to a speech therapist (frequency with which an adult with down syndrome (and his/her relatives) visits a speech therapist)
- O Visit to a psychologist (frequency with which an adult with down syndrome (and his/her relatives) visits a psychologist)
- O Thyroid screening
- O Coeliac disease screening
- O Palliative care
- O Other elements / topics, namely: ……
- O I don't know

[IF answer to 11a. was "very important", "important", or "neutral":]

11d. Please explain your answers concerning a future guideline for health care for adults with Down syndrome. (optional)
…

12. Health care for adults (18 years and older) with Down syndrome:

12a. Do you think that the set of indicators should include indicators reflecting the use of additional guidelines, protocols and / or quality standards?
For example: screening lists (like "Health Watch"), guidelines in your field of expertise, or from the Dutch quality framework for care for people with intellectual disabilities, or other)

	Very important	Important	Neutral	Not that important	Not important at all	I don't know
The indicator set should be able to measure adherence to additional guidelines / protocols / quality standards	O	O	O	O	O	O

[IF answer to 12a. was "very important", "important", or "neutral":]

12b. Which additional guidelines / protocols / quality standards?
(For example: guidelines in your field of expertise, or from the Dutch quality framework for care for people with intellectual disabilities, or other)
…

12c. Please explain (optional)
…

13. Health care for adults (18 years and older) with Down syndrome:

13a. Which quality domains or topics should be reflected in the set of indicators?

	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
Clinical outcomes (for example: improved serology, heart function, BMI)	O	O	O	O	O	…
Outcomes relevant for the patient (e.g. quality of life, daily functioning, participation)	O	O	O	O	O	…
Coordination *within* an organisation or department (for example: presence of a coordination, multidisciplinary consultation)	O	O	O	O	O	…
Coordination *between* health care professionals of different organisations and sectors (for example: between health care professionals in primary care, secondary care, or social care)	O	O	O	O	O	…
Transition 18- to 18+ (transition / transfer from paediatric to adult health care)	O	O	O	O	O	…
Findability (information on available health care providers and their differences)	O	O	O	O	O	…
Accessibility (for example: ease of making appointments, waiting time, geographical location)	O	O	O	O	O	…
Expertise (for example knowledge present among health care professionals)	O	O	O	O	O	…
Person-centeredness (for example: patient-professional relation, communication, taking into account life phase and preferences of patient and parents)	O	O	O	O	O	…
Equality	O	O	O	O	O	…

13b. Which additional quality domains or topics should be reflected in the set of indicators? (in other words, do you miss quality domains or topics in the above list and how important do you consider them to be?

PLEASE NOTE: this question (still) concerns adults with Down syndrome.

(If you do not want to add quality domains or topics, please click "next").

Add quality domains or topics below	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…

The following questions concern health care for both children and adults with Down syndrome.

14. Indicators for health care for people with Down syndrome in practice.

14a. What is the most important purpose of a set of indicators?

Indicators have to provide information in order to…	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
…map available health care, providing people with Down syndrome and their family with information on where to find good health care.	O	O	O	O	O	…
…minimalise geographical differences in health care supply and quality, leading to an even distribution of quality across health care providers in the Netherlands.	O	O	O	O	O	…
…improve health care for people with Down syndrome in the Netherlands.	O	O	O	O	O	…
…improve health care for people with Down syndrome provided in my hospital / department / practice / organisation.	O	O	O	O	O	…
…inform development of guideline(s) for health care for people with Down syndrome.	O	O	O	O	O	…
…provide input for health care purchasing by health insurers	O	O	O	O	O	…
…inspect and control quality and safety of health care for people with Down syndrome (by the national inspectorate)	O	O	O	O	O	…
…inform (national) policy concerning health care for people with Down syndrome	O	O	O	O	O	…
…enable scientific research.	O	O	O	O	O	…

14b. Please indicate whether you think the set of indicators should serve additional purposes, and indicate the importance of these purposes. (in other words, do you miss purposes in the above list?)

(If you do not want to add purposes, please click "next").

Add purposes below	Very important	Important	Neutral	Not that important	Not important at all	Explain your answer (optional)
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…
…	O	O	O	O	O	…

The following questions (still) concern health care for both children and adults with Down syndrome.

15. Indicators for health care for people with Down syndrome in practice.

15a. Which guideline(s) are you using in your work with people with Down syndrome?
…

15b. Which indicators are you using in your work with people with Down syndrome? Or in what way is quality of health care being monitored by your organisation?
…

The following questions (still) concern health care for both children and adults with Down syndrome.

16. Information source(s) for indicators.
Information from different sources may be used to provide insight into quality of health care, such as: information obtained from electronic medical records and from questionnaires for patients. Perhaps, you also register information in an electronic medical record, or your organisation asks patients to fill out a questionnaire on experienced care.

16a. On which health care elements / disciplines does your organisation / practice / department collect quality information?
(more answers possible)

- O Downteam for children
- O Downteam for adults
- O Physiotherapy
- O Speech therapy
- O Dietetics
- O Occupation therapy
- O Podiatry
- O Youth care (municipal health service: screening and vaccinations)
- O Dental hygiene
- O Regular dentistry (primary care)
- O Paediatric / specialised dentist
- O General practice

- O Other health care (providers), namely: ….
- O My organisation / practice / department does not collect any quality information on health care for people with Down syndrome.
- O Not applicable, I am (currently) not employed by a health care organisation providing health care to people with Down syndrome.

16b. Please explain your answer (optional)
…

16c. On which quality domains / topics does your organisation / practice / department collect quality information?
(more answers possible)

- O On topics mentioned in the guideline(s) I use
- O Transition from paediatric to adult health care
- O Clinical outcomes (for example: improved serology, heart function, BMI).
- O Outcomes relevant for the patient (e.g. quality of life, daily functioning, participation)
- O Coordination *within* an organisation or department (for example: presence of a coordination, multidisciplinary consultation)
- O Coordination *between* health care professionals of different organisations and sectors (for example: between health care professionals in primary care, secondary care, or social care)
- O Findability (information on available health care providers and their differences)
- O Accessibility (for example: ease of making appointments, waiting time, geographical location)
- O Expertise of health care professionals
- O Person-centeredness (for example: patient-professional relation, communication, taking into account life phase and preferences of patient and parents)
- O Equality
- O Other quality domains / topics, namely: …
- O My organisation / practice / department does not collect any quality information on health care for people with Down syndrome.
- O Not applicable, I am (currently) not employed by a health care organisation providing health care to people with Down syndrome.

16b. Please explain your answer (optional)
…

17. Information source(s) for indicators.
For this set of indicators we aim to use information that is already being registered or available as much as possible. However, a possible outcome of this study may be that additional information (not yet being registered / available) is needed for the set of indicators.

17a. Do you think your colleagues (having the same profession) will be willing to register additional information (next to what is already being registered)?

- o No, I don't think so.
- o Only if this entails just clicking a few extra boxes in the registration system.
- o Yes, I think they will.

17b. Please explain your answer (for example: concerning your ideas on how to collect information)
(optional)
…

18. The final set of indicators

The development of a set of indicators that is applicable to all Downteams in the Netherlands within this research project is a feasible exercise, because Downteams form a delineated part of health care for people with Down syndrome. Preferably, we would like to develop indicators for all health care providers involved in health care for people with Down syndrome. However, we will face several issues, such as:

- Should the set of indicators be applicable to both health care professionals with frequent and with occasional contact with people with Down syndrome?
  - If the set would only be applicable to health care professionals with frequent contact with people with Down syndrome, how to define and identify these professionals?
  - If the set would also be applicable to health care professionals with only occasional contact with people with Down syndrome, is it fair and valid to measure quality of provided care by these professionals? (for example: is it fair to expect each general practitioner to have expertise on Down syndrome?)
- On which level should the set of indicators provide information? (for example: on the level of health care professionals, organisations, departments, or on regional or municipal level)
- Where is the needed information to be found? (for example: if the set would be applicable to a region, how to make sure that health care providers within that region provide the needed data and who will be responsible to collect and manage these data?)

Please reflect on the above issues. What would you advise us to do?
…

19. Are there additional topics you would like to share with us concerning the set of indicators for health care for people with Down syndrome?
…

20. Are there other issues concerning this study (this, and the following questionnaires) you would like to share with us?
…

This is the end of this questionnaire.

By clicking "next" you will complete the questionnaire and you will not be able to adapt your answers any more.

(if you would like to change your answers, please click "back" to go to the answer(s) you would like to change. You will not lose any given answers)

Round 2

Dear participant,
Thank you very much for your willingness to take part in this study. This study concerns quality indicators for measuring quality of health care for people with Down syndrome. The aim of this study is to identify potential quality indicators and to reveal how these indicators may be used in practice.

The study involves four rounds: the first one, which is completed, and round two, three and four, which will take place today. Today, you will receive three questionnaires: at 10.00am (the current one), around 01.00pm, and around 15.30pm.
Please complete the current questionnaire (round 2) by 11.00am.

You gave informed consent for participation in this study. Please note that participation is on a voluntary basis. You are free to withdraw from the study at any moment, without an explicit reason.

Please do not hesitate to contact us if you experience any problems [phone number and e-mail address of first author].
Good luck!

Kind regards,
[names of the authors]

Please click "next" to start the questionnaire.

1. Not all participants were able to fill out the first questionnaire (round 1).
Did you complete the first questionnaire?
- o Yes
- o No

[IF answer to question 1 was "No"]

2. What is your age? (scroll down menu 20-90)

3. What is your gender?
- o Male
- o Female

4. What is your current professional position?
If you are currently not employed, please mention this in your answer and indicate the position you have had for the longest period.
…

5. (approximately) how long did you work / have you been working in this position?
If you are currently not employed, please indicate how long you worked in the position indicated in the former question.
…

6a. Professionally, how often are you in contact with people with Down syndrome (children and/or adults)?
If you are currently not employed, please indicate how often you were professionally in contact with people with Down syndrome in the position mentioned in the former question.

- o (almost) daily
- o Weekly
- o Monthly
- o Biannually
- o Annually
- o Less than once a year

6b. Explanation related to your profession and/or your contact with people with Down syndrome (questions 3-5)
(optional)
…

Indicators
Based on literature, existing guidelines, previous input of health care professionals and interviews with people with Down syndrome, their parents, and support staff, we identified relevant elements of quality of health care for people with Down syndrome. The number of elements appeared to be large.

"An indicator is a 'measurable element of practice performance (…) that can be used to assess the quality, and hence change in quality, of care provided" (Lawrence et al., 1997). An indicator is a signalling agent: it is not a direct measure of quality, but indicates a certain aspect of healthcare provision, which may be reason for further investigation. (Handleiding indicatorenontwikkeling, Kennisinstituut Medisch Specialisten, 2013)

Preferably, a small number of indicators provides as much information as possible. In other words, we strive to obtain a good impression of quality of health care for people with Down syndrome using only a few indicators. An important reason for this is to limit administrative burden. Therefore, this study aims to select indicators that best reflect health care quality, and at the same time, lead to the least administrative burden.

This study
With the current and the following questionnaires, we aim to reveal the topics that are, according to you, relevant for a set of indicators for health care for people with Down syndrome. We would also like to get insight into your ideas about how such a set should or could be used in practice. In the questionnaires, the following themes will be addressed:

- Aim and execution of this study
- Quality of health care for people with Down syndrome
- Purposes and use of a set of indicators
- Quality domains the set should focus on
- Health care disciplines the set should be covering.

Aim and execution of this study

7a. Please indicate the extent to which you agree with the following proposition:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
The questionnaires (this study) have to be used to identify the relevant topics of the set of indicators. The actual development of the indicators ("How to measure quality on these topics?") has to be done by the researchers and relevant stakeholders.	O	O	O	O	O

7b. According to you, who should be involved in the development of the set of indicators?
…

7c. Would you be willing to be involved in the further development of the set of indicators?
- o Yes
- o No
- o May be (depending on the time investment needed, the planning of the development, etc.)

8. Normally, before a set of indicators is being finalised, a concept version of such a set is presented to experts for consultation.
Which experts do you think should be consulted?
…

9. Finally, the final set of indicators will be presented to the Dutch Health care Institute, in order to register the set in a national database of quality instruments for the Dutch health care. The institute demands that quality instruments are approved by representative of at least patients / clients, health care providers, and health insurers.

9a. Representatives of which patient / client groups or organisations do you think should be involved?
…

9b. Representatives of which health care providers do you think should be involved?
…

9c. Representatives of which health insurers do you think should be involved?
…

9d. Representatives of which other groups should be involved? (optional)
…

10a. According to you, what should be the maximum number of indicators in the set? (scroll down menu 1-75)

10b. Please explain your answer. (optional)
…

Quality of health care for people with Down syndrome

11. The following propositions concern quality of health care for people with Down syndrome.

11a. Please indicate the extent to which you agree with the following propositions:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
1. An up-to-date multidisciplinary guideline is crucial for quality in health care for people with Down syndrome.	O	O	O	O	O
2. Guidelines should always contain quality indicators measuring quality of health care.	O	O	O	O	O
3. In theory, guidelines are nice, but in practice, they are barely applicable.	O	O	O	O	O
4. Indicators for health care for people with Down syndrome should be based on an up-to-date multidisciplinary guideline.	O	O	O	O	O
5. Indicators do not contribute to quality of health care for people with Down syndrome.	O	O	O	O	O
6. Health care for children with Down syndrome should strive to enable parents / relatives to decide about health care for their child / relative with Down syndrome.	O	O	O	O	O
7. Health care for adults with Down syndrome should enable them / their relatives to decide about health care they receive.	O	O	O	O	O
8. The purpose of health care for people with Down syndrome is to improve quality of life of people with Down syndrome.	O	O	O	O	O
9. Quality of health care for people with Down syndrome is highly dependent on the presence of a health care professional who coordinates the large number of disciplines involved.	O	O	O	O	O
10. High quality in health care for children with Down syndrome can only be achieved if health care professionals collaborate in multidisciplinary teams on a permanent basis, which is the case in existing Downteams in the Netherlands.	O	O	O	O	O
11. High quality in health care for adults with Down syndrome can only be achieved if health care professionals collaborate in multidisciplinary teams on a permanent basis, which is the case in existing Downteams in the Netherlands.	O	O	O	O	O
12. Downteams do not bring any added value to existing clinics for people with intellectual disabilities, having expertise in several syndromes and contact with several health care professionals.	O	O	O	O	O

	Totally agree	Agree	Neutral	Disagree	Totally disagree
13. Quality of health care for people with Down syndrome is highly dependent on the competences of individual health care professionals.	O	O	O	O	O
14. Health care professionals not having contact with people with Down syndrome on a regular basis cannot be expected to have much expertise in Down syndrome.	O	O	O	O	O
15. General practitioners and other health care professionals having only occasional contact with people with Down syndrome do not need to have much knowledge on Down syndrome, as long as they know where to find information, or to which health care professionals they can make referrals.	O	O	O	O	O

11b. Please explain your answers regarding the above propositions. (optional)
…

Purposes and use of the set of indicators

Participants in the previous round considered the following purposes for the set of indicators (very) important:
- Providing insight into available health care (providers)
- Minimalise geographical differences in health care supply and quality, leading to an even distribution of quality across health care providers in the Netherlands.
- Improve health care for people with Down syndrome in the Netherlands.
- Inform development of guideline(s) for health care for people with Down syndrome.
- Inspect and control quality and safety of health care for people with Down syndrome (by the national inspectorate)

The opinions of the participants concerning the following purposes were diverse:
- Improve health care for people with Down syndrome provided in my hospital / department / practice / organisation.

Participants considered the following purposes not important:
- Provide input for health care purchasing by health insurers
- Inform (national) policy concerning health care for people with Down syndrome
- Enable scientific research.

It is remarkable that opinions of participants in the previous round concerning "Improve health care for people with Down syndrome provided in my hospital / department / practice / organisation" were very divided, given that they indicated the other propositions on health care improvement as being important.

12. In order to find out more about your opinion and its details, and to provide you with the opportunity to revise your previous answers, the following propositions are presented to you.

12a. Please indicate the extent to which you agree with the following propositions:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
1. The set of indicators has to provide information in order to inform development of guideline(s) for health care for people with Down syndrome.	O	O	O	O	O
2. The set of indicators has to provide information in order to improve health care for people with Down syndrome provided in my hospital / department / practice / organisation.	O	O	O	O	O
3. The set of indicators has to provide insight into which hospital / department / practice / organisation provides good health care and which does not.	O	O	O	O	O

In order to obtain more insight into your opinion regarding "Providing insight into available health care (providers)", we prepared the following propositions.

12b. Please indicate the extent to which you agree with the following propositions:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
4. The set of indicators has to provide people with DS, their parents, and support staff with information on where to find suitable health care.	O	O	O	O	O
5. The set of indicators has to provide health care professionals with information on where to find good health care for people with Down syndrome (for making referrals).	O	O	O	O	O
6. The set of indicators has to provide people with DS, their parents, and support staff with information on their rights in health care.					
7. The set of indicators has to provide people with DS, their parents, and support staff with information on available NON-medical care (such as family support).	O	O	O	O	O
8. The set of indicators has to provide health care professionals with information on available NON-medical care (such as family support).	O	O	O	O	O

12c. We would also like to present the following propositions to you (again).

The set of indicators should provide information suitable as input or guidance for …	Very important	Important	Neutral	Not that important	Not important at all
9. … health care purchasing by health insurers	O	O	O	O	O
10. … inspection and control of quality and safety (by the national inspectorate)	O	O	O	O	O
11. … (national) policy	O	O	O	O	O
12. … scientific research	O	O	O	O	O

Do you miss purposes? Would you like to explain your answers?

12d. Please indicate whether you have missed purposes in the above list. You may also want to explain your answers (concerning purposes of the set of indicators), please do so below. (optional)

I would like to add the following purposes:
…

I would like to explain my answers:
…

Quality domains
In the previous round, participants indicated which quality domains or topics they thought the set of indicators should cover, and which domains or topics the considered less important.

Participants in the previous round considered the following quality domains important for the set of indicators:

Quality domain	Participants' comments
Clinical outcomes (for example: improved serology, heart function, BMI)	*Important to monitor.* *Does not necessarily indicate quality.*
Outcomes relevant for the patient (e.g. quality of life, daily functioning, participation)	*Is very important for a happy feeling.* *Does not necessarily indicate quality of provided care, is also influenced by other factors.*
Coordination *within* an organisation or department	*Collaboration is very important.*
Coordination *between* health care professionals of different organisations and sectors	*Important for making the right, and timely, referrals.*
Transition 18- to 18+	*Currently, this does not run smoothly, there is a 'gap' after 18.* *It is important that information does not need to be gathered anew.*

Quality domain	Participants' comments
Expertise	*Health care professionals need to know where expertise is to be found.*
Person-centeredness	*But consider privacy.*

Participants considered the following quality domains less important:

Quality domain	Participants' comments
Findability	*It is important for getting the right care.* *May be the purpose of a potential set of indicators.*
Accessibility	*Important, but difficult to improve.*
Equality	*Equality is taken for granted*

The following quality domains were added:

Quality domain	Participants' comments
Empowerment, self-management	*Also consider explicit involvement of legal representative of the person with Down syndrome.*
Participation in society	*Quality of housing*
Religion, spirituality	*Leisure, work, school, daily activities*

13. The above topics / domains are presented to you again, with a few adaptations (based on the comments given by the participants).

13a. Please indicate how important you consider the following quality domains for the set of indicators.

The set of indicators should provide insight into…	Very important	Important	Neutral	Not that important	Not important at all
… how people with DS in the Netherlands are doing (for instance by providing information on prevalence of disease, overweight, quality of life, functioning or participation in society)	O	O	O	O	O
… adherence to guidelines (for example by monitoring whether screenings mentioned in guidelines are carried out in time).	O	O	O	O	O
… coordination and collaboration within AND between organisations.	O	O	O	O	O
… transition from paediatric health care (until the age of 18) to adult health care (starting at the age of 18).	O	O	O	O	O
… effectiveness (such as effect of interventions, expertise of health care professionals, timely recognition of health problems).	O	O	O	O	O

The set of indicators should provide insight into…	Very important	Important	Neutral	Not that important	Not important at all
… person-centeredness (such as interaction between health care professionals and person with Down syndrome, health care tailored to the desires and possibilities of people with Down syndrome).	O	O	O	O	O
… safety of provided health care for people with Down syndrome.	O	O	O	O	O

Do you miss quality domains or topics? Would you like to explain your answers?

13b. Please indicate whether you have missed quality domains or topics in the above list. You may also want to explain your answers (concerning quality domains or topics the set of indicators should cover), please do so below. (optional)

Please note: this question only concerns quality domains / topics. Health care elements and disciplines to be covered by the set of indicators will be addressed further on in the questionnaire.

I would like to add the following quality domains / topics:
…

I would like to explain my answers:
…

Structure, process and outcome indicators

Quality of health care is often described in terms of health care structure, care processes, and health outcomes. Accordingly, indicators can be grouped into structure, process and outcome indicators. By *structure* of health care we mean the health care system: availability of facilities and qualified staff, rules and regulations, protocols, and financial means (including health insurance). Care *processes* are: all actions taking place between patients and health care professionals, both technical interventions (such as measuring blood pressure), and interactions between professional and patient (such as communication). An often-used measure for quality of health care is adherence to guidelines. *Outcomes* of health care reflect the result of provided health care: whether the patient's situation has improved or not. Examples of outcome measures are presence/absence of disease, increase/decrease of complaints, quality of life. An outcome indicator is a measure for the total care path, including the processes and structures, which contributed to the outcome.

What type of indicators do you think the set should include? (structure, process, outcome indicators?)
Below you can indicate the ideal proportion of structure, process, and outcome indicators in the set. You can indicate this by dividing 60 points over the three types of indicators.

For example: If you think the set of indicators should merely consist of process indicators, you should allocate 60 points to 'Process indicators'. If you think that the number of structure, process and outcome indicators should be equal, you should allocate 20 point to each type of indicator.

14. You may divide the points however you like, if only the sum of the points is 60.
Structure (scroll down: 0-60)
Process (scroll down: 0-60)
Outcome (scroll down: 0-60)
Total (automated sum of scores)

Health care elements or disciplines the set of indicators should cover

In the previous round, we asked participants which health care elements or disciplines they considered important to be covered by the set of indicators. An overview of elements or disciplines that were considered important and the ones that were considered less important is presented below. We also provide insight into health care elements or disciplines that were added by the participants in the previous round. The left column concerns health care for children with Down syndrome, the right column concerns health care for adults with Down syndrome. We also provide a summary of given comments.

The following health care elements / disciplines were considered important according to the participants in the previous round:

Health care elements / disciplines for CHILDREN with Down syndrome	Health care elements / disciplines for ADULTS with Down syndrome
Downteam *("should be available for all children with Down syndrome"; "health care professionals need to collaborate"; "contributes to efficiency")*	Downteam *("Much room for improvements")*
Paediatrician *("essential as part of a Downteam, but also as 'mono-'discipline")*	ID physician *("the number of ID physicians is too low")*
Physiotherapy *("high prevalence of movement problems"; "plays an important advisory role")*	
Speech therapy *("has a positive influence on other health problems")*	
Dietetics *("Important to acquire a good eating pattern and to prevent overweight")*	Dietetics
Psychological care *("should be available to everyone"; "there is too less uniformity in psychological care")*	Psychological care
General practitioner *("primary contact in health care"; "has little knowledge on Down syndrome")*	General practitioner *("primary contact in health care"; "has little knowledge on Down syndrome")*
ENT physician	Palliative care *("it is important to address this in time"; "including dementia and functional decline")*
Thyroid screening	
Heart echo	

The following health care elements / disciplines were considered less important according to the participants:

Health care elements / disciplines for CHILDREN with Down syndrome	Health care elements / disciplines for ADULTS with Down syndrome
Occupational therapy *("brings an added value as compared to physiotherapy and is deployed too little")*	Occupational therapy *("important for general daily activities are important")*
Podiatrist *("high prevalence of feet and walking problems"; "Someone should coordinate aids and treatment")*	Podiatrist *("high prevalence of feet and walking problems"; "Someone should coordinate aids and treatment"; "pain, inactivity, overweight, and complaints are often caused by problems related to feet/walking/shoes")*
Dermatology *("skin problems generally start from puberty, and often receive insufficient attention")*	Dermatology *("more important that for children")*
Youth care *("important for vaccinations, coordination and integration at school")*	Physiotherapy *("only when needed")*
Dental care *("Dental hygiene, regular dental care and specialised dental care are important, but regular specialised care is desirable"; "had impact on other health problems")*	Dental care *("Dental hygiene, regular dental care and specialised dental care are important, but regular specialised care is desirable"; "had impact on other health problems")*
Coeliac disease screening	Coeliac disease screening
	Thyroid screening
	Speech therapy *("Improvements are always possible"; "communication, speech, and language are important prerequisites for functioning")*
	ENT-physician
	Ophthalmology / orthoptist

The following health care elements / disciplines were added by the participants:

Health care elements / disciplines for CHILDREN with Down syndrome	Health care elements / disciplines for ADULTS with Down syndrome
(paediatric) cardiology	
ID physician *("in case of complex behaviours or sleeping problems")*	
Audiology / hearing screening *("an ENT-physician is not always needed")*	Audiology / hearing screening *("an ENT-physician is not always needed")*
Paediatric psychiatry	Psychiatry *("related to depression/anxiety"; "where to find suitable care?")*
Orthopaedics *("screening of knees and hips")*	Orthopaedics *("screening of feet, knees, and hips")*
Paediatric rehabilitation specialist	
Physical activity professional *("for developing a healthy exercise pattern")*	
Multidisciplinary sleeping research team *("related to sleeping apnoea")*	Multidisciplinary sleeping research team *("related to sleeping apnoea")*

Health care elements / disciplines for CHILDREN with Down syndrome	Health care elements / disciplines for ADULTS with Down syndrome
Ophthalmology / orthoptist *("important for daily functioning and vision development")*	Optometrist *("complementary to Ophthalmology / orthoptist")*
Centre of expertise for blind people	
Specialised (ID) nurse	
Organisations for people with ID	
Support staff *("for children not living with their parents")*	
Diabetes screening	Diabetes screening
Sexuality / puberty / contraception	
Family support *("professional guidance concerning 'Early intervention' / coping with a child with Down syndrome / choosing schools / respite care / local community")*	Case manager / mentor *("has to coordinate care, because parents of adults with Down syndrome are not able to do that anymore")*

15. Which health care disciplines / elements should be covered by the set of indicators?
On the previous page, we presented health care disciplines / elements that, according to the participants in the previous round, should be covered by the set of indicators. Here we present these disciplines / elements again, including newly added disciplines / elements. Please keep in mind that the set of indicators should be as compact and concise as possible. We ask you to indicate only those elements / disciplines as "(very) important" if you think these are crucial for obtaining insight into health care for people with Down syndrome.

15a. Which elements of health care should be reflected in the set of indicators?

Please indicate this for children (left column) and for adults (right column).

	CHILDREN					ADULTS				
	Very important	Important	Neutral	Not that important	Not important at all	Very important	Important	Neutral	Not that important	Not important at all
Downteam (a coordinated team of collaborating multidisciplinary health care providers collaborating for children or adults with down syndrome)	O	O	O	O	O	O	O	O	O	O
Paediatrician	O	O	O	O	O	O	O	O	O	O
ID physician	O	O	O	O	O	O	O	O	O	O
Physiotherapy	O	O	O	O	O	O	O	O	O	O
Speech therapist	O	O	O	O	O	O	O	O	O	O
Dietician	O	O	O	O	O	O	O	O	O	O
Occupational therapy	O	O	O	O	O	O	O	O	O	O
Podiatrist	O	O	O	O	O	O	O	O	O	O

	CHILDREN					ADULTS				
	Very important	Important	Neutral	Not that important	Not important at all	Very important	Important	Neutral	Not that important	Not important at all
Dermatology	O	O	O	O	O	O	O	O	O	O
Mental health care	O	O	O	O	O	O	O	O	O	O
Youth health care (municipal health service)	O	O	O	O	O	O	O	O	O	O
Dental care: Dental hygienist, regular dentist (primary care), specialised dentist, and orthodontist	O	O	O	O	O	O	O	O	O	O
General practitioner	O	O	O	O	O	O	O	O	O	O
Care for dementia and functional decline	O	O	O	O	O	O	O	O	O	O
Palliative care	O	O	O	O	O	O	O	O	O	O
Cardiology	O	O	O	O	O	O	O	O	O	O
Rehabilitation	O	O	O	O	O	O	O	O	O	O
Orthopaedics	O	O	O	O	O	O	O	O	O	O
Physical activity professional	O	O	O	O	O	O	O	O	O	O
ENT-specialist	O	O	O	O	O	O	O	O	O	O
Audiology / hearing screening	O	O	O	O	O	O	O	O	O	O
Ophthalmology / orthoptist	O	O	O	O	O	O	O	O	O	O
Optometrist	O	O	O	O	O	O	O	O	O	O
Centre of expertise for blind people	O	O	O	O	O	O	O	O	O	O
Multidisciplinary sleeping research team	O	O	O	O	O	O	O	O	O	O
Screening for coeliac disease	O	O	O	O	O	O	O	O	O	O
Screening for thyroid disease	O	O	O	O	O	O	O	O	O	O
Screening for diabetes	O	O	O	O	O	O	O	O	O	O
ID specialised nurse / practice nurse	O	O	O	O	O	O	O	O	O	O
Support staff in living facilities	O	O	O	O	O	O	O	O	O	O
Professional family support	O	O	O	O	O	O	O	O	O	O
Case manager / mentor	O	O	O	O	O	O	O	O	O	O

Do you miss health care disciplines or elements? Would you like to explain your answers?

15b. Please indicate whether you have missed health care elements or disciplines in the above list. You may also want to explain your answers (concerning health care elements or disciplines the set of indicators should cover), please do so below. (optional)

I would like to add the following health care elements or disciplines:
…

I would like to explain my answers:
…

16. The following propositions also concern the health care elements or disciplines that should be covered by the set of indicators.

16a. Please indicate the extent to which you agree with the following propositions:
(and explain your answers if you like)

	Totally agree	Agree	Neutral	Disagree	Totally disagree
Health care elements or disciplines are more important for the set of indicators when more people with Down syndrome need them.	O	O	O	O	O
Health care elements or disciplines are more important for the set of indicators when they contribute more to quality of life of people with Down syndrome.	O	O	O	O	O
Health care elements or disciplines are more important for the set of indicators when there are more providing professionals.	O	O	O	O	O
Health care elements or disciplines are more important for the set of indicators when there are more doubts about the quality of the element / discipline.	O	O	O	O	O
Health care for people with Down syndrome is multidisciplinary. Therefore, the set of indicators should cover all disciplines involved in health care for people with Down syndrome.	O	O	O	O	O
Relevance of health care elements / disciplines depends on the life phase of a person with Down syndrome. Accordingly, each life phase needs different indicators.	O	O	O	O	O

16b. Please explain you answers to the above propositions. (optional)
…

This is the end of questionnaire 2.

Be aware: after clicking "next", you are not able to adapt your answers anymore!

(if you would like to change your answers, please click "back" to go to the answer(s) you would like to change. You will not lose any given answers)

Round 3

Dear participant,

This morning you completed the questionnaire of round 2 of the study. Thanks! In the current questionnaire we present themes similar to the ones in the previous questionnaire(s). In the current questionnaire, the themes on which no consensus was achieved among the participants in the previous round are addressed, and themes on which more detailed information is needed. Please complete the current questionnaire (round 3) by 2.15 pm.

You will receive the next (and last) questionnaire at 3.30 pm.
You gave informed consent for participation in this study. Please note that participation is on a voluntary basis. You are free to withdraw from the study at any moment, without an explicit reason.

Please do not hesitate to contact us if you experience any problems [phone number and e-mail address of first author].
Good luck!

Kind regards,
[names of the authors]

Please click "next" to start the questionnaire.

Aims and execution of the study
1. A set of indicators should not be developed without involving:
- O Clients (people with Down syndrome)
- O Parents / relatives
- O Support staff or health care professionals
- O Health insurers

Purposes and use of the set of indicators
Based on the outcomes of the previous rounds, we formulated the following questions:
2. How could the indicators provide information that could be used by people with Down syndrome and their relatives for choosing suitable health care, without naming health care professionals?
…

3. How could the indicators provide information that could be used by health care organisations or professionals to improve provided care, without naming health care professionals and organisations?
…

4. From the previous round, it appeared that participants considered "providing information that enables improvements in care provision by my organisation" an important purpose of the set of indicators.

How do you think the information provided by the set of indicators should be used?
4a. Please indicate the extent to which you agree with the following propositions and, if desired, please add purposes of the set of indicators.

	Totally agree	Agree	Neutral	Disagree	Totally disagree
16. Information provided by the indicators should be suitable for short-term evaluations on the level of patients/clients.	O	O	O	O	O
17. Information provided by the indicators should be suitable for interdisciplinary evaluation.	O	O	O	O	O
18. Information provided by the indicators should be suitable for clinical-epidemiological research.	O	O	O	O	O
19. Information provided by the indicators should be suitable as input for adjusting protocols.	O	O	O	O	O

4b. Which additional purposes, concerning use of indicators for health care improvements, would you like to add? Please indicate below: (optional)
…

5. From the previous round, it appeared that participants considered "providing information as input for health care purchasing by health insurers" and "providing information as input for inspection and review" important purposes for the set of indicators. More detailed purposes are formulated below.
5a. Please indicate the extent to which you agree with the following propositions and, if desired, please add purposes for the indicators.

	Totally agree	Agree	Neutral	Disagree	Totally disagree
1. Information provided by the indicators should be suitable as input for negotiations about health care purchasing.	O	O	O	O	O
2. Information provided by the indicators should be suitable as input for contracting health care providers by health insurers.	O	O	O	O	O
3. Information provided by the indicators should be suitable for assessment of performance of professionals and rewards.	O	O	O	O	O
4. Information provided by the indicators should be suitable as input for inspection by the national inspectorate.	O	O	O	O	O
5. Information provided by the indicators should be suitable as input for review and control by the supervisory board.	O	O	O	O	O

5b. Which additional purposes, concerning use of indicators by health insurers or for control, would you like to add? Please indicate below: (optional)
…

6. Detail and level of the set of indicators
6a. Please indicate the extent to which you agree with the following propositions.

	Totally agree	Agree	Neutral	Disagree	Totally disagree
1. The set of indicators should be modular in order to allow users to choose which information they would like to register.	O	O	O	O	O
2. Next to a general set containing indicators of all disciplines, there should be an elaborated set per discipline.	O	O	O	O	O
3. The set should provide quality information on organisational / departmental level.	O	O	O	O	O
4. The set should provide quality information on the level of individual professionals.	O	O	O	O	O
5. There should be a quality mark for professionals / organisations specialised in Down syndrome.	O	O	O	O	O
6. Health care organisations / departments should publish quality information on their websites.	O	O	O	O	O
7. The set of indicators should only include health care professionals with frequent contact with people with DS.	O	O	O	O	O
8. Joining the set of indicators should be voluntary, and could be an opportunity for health care providers to display their expertise.	O	O	O	O	O

6b. Please explain your answers concerning the above propositions. (optional)
…

7. From the previous round, it appeared that participants considered the following quality domains and topics important to be covered by the set of indicators:
- "How people with Down syndrome in the Netherlands are doing"
- "Adherence to guidelines"
- "Coordination and collaboration within and between organisations"
- "Transition from paediatric health care to adult health care"
- "Effectiveness"
- "Person-centeredness"
- "Safety".

We now elaborate on these issues and ask you to indicate which (more specific) quality domains and topics you think should be covered by the set of indicators.

Concerning "How people with Down syndrome in the Netherlands are doing"
7a. How important do you think it is that the set provides insight into…

	Very important	Important	Neutral	Not that important	Not important at all
… whether burden for the social environment is taken into account	O	O	O	O	O
… whether caregivers have the feeling to have control over the health care of the person with Down syndrome.	O	O	O	O	O
… autonomy of the person with Down syndrome.	O	O	O	O	O
… daily functioning of a person with Down syndrome.	O	O	O	O	O
… quality of life of a person with Down syndrome.	O	O	O	O	O
… participation in society of a person with Down syndrome (for instance: in relation to work, school, leisure, daily activity centres).	O	O	O	O	O
… personal development of a person with Down syndrome, such as motor skills, and sensory, cognitive and speech development.	O	O	O	O	O
… experienced health problems, such as pain and fatigue.	O	O	O	O	O
… measurable physical health (e.g. BMI, blood tests).	O	O	O	O	O
… physical health, as experienced by the person with Down syndrome.	O	O	O	O	O
… mental health, as experienced by the person with Down syndrome.	O	O	O	O	O

7b. Please explain your answers to the above propositions and add topics: (optional)
Please note: health care elements / disciplines to be covered by the set of indicators will follow later on in the questionnaire.
….

Concerning "Coordination and collaboration within and between organisations"
7c. How important do you think it is that the set provides insight into…

	Very important	Important	Neutral	Not that important	Not important at all
… mutual collaboration among health care professionals.	O	O	O	O	O
… collaboration between health care professionals and parents / relatives / mentors.	O	O	O	O	O
… mutual agreements among health care providers about tasks and responsibilities.	O	O	O	O	O
… agreements between health care professionals and parents / relatives / mentors about tasks and responsibilities.	O	O	O	O	O
… coordination within organisations or departments.	O	O	O	O	O

… collaboration and knowledge sharing between professionals from different organisations and disciplines.	O	O	O	O	O

Concerning "Transition from paediatric health care to adult health care"
7d. How important do you think it is that the set provides insight into…

	Very important	Important	Neutral	Not that important	Not important at all
…the presence of a transition protocol	O	O	O	O	O
… the way in which transition takes place.	O	O	O	O	O

7e. Please explain your answers to the above propositions and add topics: (optional)
Please note: health care elements / disciplines to be covered by the set of indicators will follow later on in the questionnaire.
….

Concerning "Person-centeredness"
7f. How important do you think it is that the set provides insight into…

	Very important	Important	Neutral	Not that important	Not important at all
… whether preferences, values, living situation etc. of the person with Down syndrome are taken into account.	O	O	O	O	O
… self-management (for example: a person with Down syndrome learns how to inject insulin).	O	O	O	O	O
… shared decision making.	O	O	O	O	O
… whether several disciplines can be visited on one day	O	O	O	O	O
… the presence of one contact person for a person with Down syndrome / caregivers parents / relatives / mentors.	O	O	O	O	O
… whether health care is nearby.	O	O	O	O	O

Concerning "Effectiveness"
7g. How important do you think it is that the set provides insight into…

	Very important	Important	Neutral	Not that important	Not important at all
… cost effectiveness.	O	O	O	O	O
… expertise of health care professionals.	O	O	O	O	O
… timely recognition of health problems.	O	O	O	O	O

7h. Please explain your answers to the above propositions and add topics: (optional)
Please note: health care elements / disciplines to be covered by the set of indicators will follow later on in the questionnaire.
….

Structure, process and outcome indicators

Quality of health care is often described in terms of health care structure, care processes, and health outcomes. Accordingly, indicators can be grouped into structure, process and outcome indicators. By *structure* of health care we mean the health care system: availability of facilities and qualified staff, rules and regulations, protocols, and financial means (including health insurance). Care *processes* are: all actions taking place between patients and health care professionals, both technical interventions (such as measuring blood pressure), and interactions between professional and patient (such as communication). An often used measure for quality of health care is adherence to guidelines. *Outcomes* of health care reflect the result of provided health care: whether the patient's situation has improved or not. Examples of outcome measures are presence/absence of disease, increase/decrease of complaints, quality of life. An outcome indicator is a measure of the total care path, including the processes and structures, which contributed to the outcome.

There was much variation in the answers of the participants in the previous rounds to the question below. Therefore, this question is presented to you again. Do you stick to your previous answer, or would you like to adjust it?

The answers in the previous round were:

	Range	Mean	Median
Structure	5-30	16.2	15
Process	10-30	19.1	20
Outcome	10-40	24.7	25

What type of indicators do you think the set should include? (structure, process, outcome indicators?)
Below you can indicate the ideal proportion of structure, process, and outcome indicators in the set. You can indicate this by dividing 60 points over the three types of indicators.

For example: If you think the set of indicators should merely consist of process indicators, you should allocate 60 points to 'Process indicators'. If you think that the number of structure, process and outcome indicators should equal, you should allocate 20 point to each type of indicator.

8a. You may divide the points however you like, if only the sum of the points is 60.
Structure (scroll down: 0-60)
Process (scroll down: 0-60)
Outcome (scroll down: 0-60)
Total (automated sum of scores)

8b. Please explain your answer regarding the type of indicators below (optional).
…

Health care elements / disciplines to be covered by the set of indicators
9a.Which health care elements / disciplines are required for (almost) all people with Down syndrome (during at least a certain period) in their lives?
Please mention the one(s) that first come in mind.
…

9b. Which health care elements / disciplines are largely contributing to quality of life of people with Down syndrome?
Please mention the one(s) that first come in mind.
…

9c. Providers of which health care discipline(s) are largely available? (Hence: people with Down syndrome and their parents / relatives may have many options for choosing a provider)
Please mention the one(s) that first come in mind.
…

9d. For which health care discipline(s) do doubts exist concerning the quality of care provided by these disciplines?
Please mention the one(s) that first come in mind.
…

This is the end of questionnaire 3.

Be aware: after clicking "next", you are not able to adapt your answers anymore!

(if you would like to change your answers, please click "back" to go to the answer(s) you would like to change. You will not lose any given answers)

Round 4

Dear participant,
Earlier today, you completed the questionnaire of round 2 and 3 of the study. Thanks! In the current questionnaire we present themes similar to the ones in the previous questionnaire(s). In the current questionnaire, themes are addressed on which no consensus was achieved among the participants in the previous round(s), and themes on which more detailed information is needed.
Please complete the current questionnaire (round 4) by tomorrow (May 31).

You gave informed consent for participation in this study. Please note that participation is on a voluntary basis. You are free to withdraw from the study at any moment, without an explicit reason.

Please do not hesitate to contact us if you experience any problems [phone number and e-mail address of first author].
Good luck!

Kind regards,
[names of the authors]

Please click "next" to start the questionnaire.

1. There was much variation in the answers of the participants in the previous rounds regarding the following proposition: "The set of indicators has to provide health care professionals with information on available NON-medical care". Therefore, we formulated the next proposition.

1a. Please indicate the extent to which you agree with the following proposition:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
A set of indicators for health care for people with Down syndrome should not contain indicators on NON-medical care (such as: daily activity centres, school, leisure time).	O	O	O	O	O

1b. Please explain your answer (optional).
…

2. From the previous round, it appeared that many participants thought that the set of indicators should consist of modules. However, round 2 did not result in consensus among participants regarding the number of indicators in the set. The answers varied from 5 to 40. Therefore, we formlated a few more questions concerning modules and the number of indicators.

2a. Please indicate the extent to which you agree with the following propositions:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
1. There should be a basic set of indicators, consisting of indicators that are relevant to all people with Down syndrome.	O	O	O	O	O
2. Next to this basic set, additional modules should be present for specific health care or patient groups.	O	O	O	O	O

[IF answer to proposition 1 is "totally agree", "agree", or "neutral":]

2b. What should be the maximum number of indicators in this basic set?
…

[IF answer to proposition 2 is "totally agree", "agree", or "neutral":]
2c. What should be the maximum number of indicators in each of these additional modules?
…

2d. Which additional modules have most priority for becoming part of the set?
…

3. Please indicate the extent to which you agree with the following propositions:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
Quality information should be public on the organisational level, but not on the provider (personal) level.	O	O	O	O	O
Health care professionals should themselves decide about public availability of quality information.	O	O	O	O	O
Privacy of professionals should be protected just as much as privacy of patients.	O	O	O	O	O
Quality of the social system of a person with Down Syndrome (including all his/her caregivers) is crucial in health care for people with Down syndrome.	O	O	O	O	O
Indicators should stimulate improvement of care and should not judge health care professionals.	O	O	O	O	O
Professionals should be obliged to register the indicators if they want to be seen as 'specialised in Down syndrome'.	O	O	O	O	O
Professionals wanting to be seen as 'specialised in Down syndrome' should be obliged to make their quality information publicly available.	O	O	O	O	O
Publishing quality information will not result in long waiting lists since most people with DS / parents will not be willing to travel far for better care.	O	O	O	O	O
Well defined outcome indicators are able to provide insight into process and structure too.	O	O	O	O	O

Practical issues related to collecting quality information

4a. Please indicate the extent to which you agree with the following propositions:

	Totally agree	Agree	Neutral	Disagree	Totally disagree
Standardisation and interoperability of electronic medical records needs to be established before quality can be measured.	O	O	O	O	O
Burden for people with Down syndrome and their caregivers should be as low as possible when measuring quality.	O	O	O	O	O
Burden for health care professionals should be as low as possible when measuring quality.	O	O	O	O	O
People with Down syndrome (and their parents/relatives) and health care professionals should both deliver information for the indicators.	O	O	O	O	O
Parents/caregivers should themselves be responsible for documenting and keeping track of needed health care for the person with Down syndrome.	O	O	O	O	O
A dialogue between health care professional and person with Down syndrome should be used as an instrument for measuring customer satisfaction.	O	O	O	O	O
An instrument measuring patient experiences or satisfaction should be suitable to be filled out by 80% of the population of people with Down syndrome by themselves.	O	O	O	O	O
When people with Down syndrome are not able to provide quality information themselves, their legal representative should decide who is eligible to provide this information.	O	O	O	O	O

4b. Please explain your answers to the above presented propositions. (optional)
…

This is the end of questionnaire 4, which is the last questionnaire of this study.

Thanks again for your willingness to take part in this study!

Be aware: after clicking "next", you are not able to adapt your answers anymore!

(if you would like to change your answers, please click "back" to go to the answer(s) you would like to change. You will not lose any given answers)

Appendix 4-II

Table 4-II Extent to which consensus was achieved among participants regarding: Purposes of QIs

	Round 1		Round 2		Round 3	
	% 1-2[a]	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*
What should be the purpose of QIs? *(How important do you consider the following purposes?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**						
Providing people with DS with information for choosing the right care	**93,8**	**1**	**78,6**	**2**		
Providing health care professionals with information for finding suitable health care providers/referrals			**82,1**	**2**		
Reducing differences in provided health care in the Netherlands	**81,3**	**2**				
Providing insight into differences between providers			60,7	2		
Improving health care quality for people with DS in the Netherlands	**96,9**	**1**				
Improving health care quality provided in own organization (round 1); provide information for doing this (round 2)[c]	65,6	2	**100**	**2**		
For short term evaluation of health care delivery on the patient level					74,1 **76[b]**	2
For interdisciplinary evaluation					**81,5**	**2**
As input for guidelines	**87,5**	**2**	71,4	2		
As input to adjust protocol					**92,6**	**2**
As input for health care purchasing by health insurers	56,3	2	42,9	3		
As input for negotiating about health care purchasing					59,3	2
As input for contracting health care providers (by insurers)					37	3
As input for performance rewards for providers					33,3	3
For inspection and review	**75**	**2**				
For inspection by the national inspectorate			**82,1**	**2**	**88,9**	**2**
For review and control by the supervisory board					**81,5**	**2**
As input for policy	65,6	2	60,7	2		
For scientific research	65,6	2	**75**	**2**		
For clinical-epidemiological research					51,9	2
Other, please add:						
Providing people with DS with information on their rights in health care			64,3	2		
A purpose of QIs should be to provide people with DS / caregivers with insight into available non-medical care			67,9	2		
A purpose of QIs should be to provide health care professionals with insight into available non-medical care.			71,4	2		

Abbreviations: QI=Quality indicator; DS=Down syndrome.
Empty fields indicate that the topic was not presented to the participants in the concerning round.

[a] %1-2 indicates the percentage of participants that had answered "very important"/"totally agree" (1) or "important"/"agree" (2). If the percentage was ≥ 75 and the median was ≤ 2, there was consensus among the participants. Consensus was indicated in **bold**.
[b] Percentage of participants that had answered "very important" (1) or "important" (2) if patient representatives are not included in analysis. The difference was only showed if exclusion of patient representatives resulted in a different conclusion regarding consensus.
[c] Formulation of purpose was different in round 1 and 2.
[d] % 4-5, indicating the percentage of participants that had answered "disagree" (4) or "totally disagree" (5).

Appendix 4-III

Table 4-III. Extent to which consensus was achieved among participants regarding: Quality domains

	Round 1				Round 2		Round 3		Round 4	
Which quality domains should be covered by the QIs? *(How important do you consider the following quality domains for QIs?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**										
	Children		**Adults**		**(Children and adults together)[b]**		**(Children and adults together)[b]**		**(Children and adults together)[b]**	
	% 1-2[a]	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*
Coordination within organisations or departments	**81,8**	**2**	**78,8**	**2**			70,4	2		
Coordination between organisations or departments	**97**	**2**	**93,9**	**2**			**100**	**1**		
Coordination within and between organisations or departments					**85,7**	**2**				
Mutual collaboration among health care providers							**88,9**	**2**		
Collaboration between health care providers and caregivers of people with DS							**88,9**	**1**		
Mutual agreements among health care providers about tasks and responsibilities							**81,5**	**2**		
Mutual agreements between health care providers and caregivers about tasks and responsibilities							74,1	2		
Quality of the social system of a person with DS (including all his/her caregivers)									69,2 **75[c]**	2
Transition from child to adult care	**100**	**2**	**97**	**1**	**96,4**	**1,5**				
The presence of a transition protocol							**85,2**	**2**		
The way in which transition takes place							**88,9**	**2**		
Whether health care is findable	69,7	2	72,7	2						
Accessibility	66,7	2	66,7	2						
Safety					**78,6**	**2**				
Equity	66,7	2	63,6	2						
Expertise	**97**	**1**	**93,9**	**2**			**85,2**	**2**		
Effectiveness					**92,9**	**1,5**				

Table 4-III. continued

	Round 1				Round 2		Round 3		Round 4	
Which quality domains should be covered by the QIs? *(How important do you consider the following quality domains for QIs?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**										
	Children		**Adults**		**(Children and adults together)[b]**		**(Children and adults together)[b]**		**(Children and adults together)[b]**	
	% 1-2[a]	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*
Cost effectiveness							51,9	2		
Timely recognition of health problems							**100**	**1**		
Person-centeredness	**87,9**	**2**	**87,9**	**1**	**85,7**	**2**				
Whether preferences, values, living situation etc. of the person with DS are taken into account							66,7	2		
Self-management							74,1	2		
Shared decision making							**96,3**	**2**		
Whether several disciplines can be visited on one day							44,4	3		
The presence of one contact person for a person with DS / caregivers							**88,9**	**2**		
Whether health care is nearby							**77,8**	**2**		
Clinical outcomes	**84,8**	**2**	**84,8**	**2**						
Outcomes relevant for the patient (e.g. quality of life, daily functioning, participation)	**97**	**1**	**97**	**1**	**82,1**	**2**				
Whether burden for the social environment is taken into account							44,4	3		
Whether caregivers have the feeling to have control over the health care of the person with DS							66,7	2		
Autonomy of the person with DS							**85,2**	**2**		
Daily functioning							**81,5**	**2**		
Quality of life							**88,9**	**1**		
Participation in society							**77,8**	**2**		
Development							**92,6**	**2**		

Table 4-III. continued

	Round 1				Round 2		Round 3		Round 4	
Which quality domains should be covered by the QIs? *(How important do you consider the following quality domains for QIs?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**										
	Children		**Adults**		**(Children and adults together)[b]**		**(Children and adults together)[b]**		**(Children and adults together)[b]**	
	% 1-2[a]	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*	*% 1-2[a]*	*median*
Experienced health problems							**96,3**	**1**		
Measurable physical health (e.g. BMI, blood tests)							**96,3**	**1**		
Experienced physical health							**92,6**	**1**		
Experienced mental health							**88,9**	**2**		
Adherence to the Dutch medical guideline for children with DS	**80,6**	**2**								
Adherence to the future Dutch medical guideline for adults with DS[d]			**86,2**	**2**						
Adherence to guidelines in general					**89,3**	**2**				

Abbreviations: QI=Quality indicator; ID=Intellectual disability; DS=Down syndrome; BMI=Body mass index.

Empty fields indicate that the topic was not presented to the participants in the concerning round.
[a] %1-2 indicates the percentage of participants that had answered "very important" (1) or "important" (2). If the percentage was ≥ 75 and the median was ≤ 2, there was consensus among the participants. Consensus was indicated in **bold**.
[b] Health care for children and adults with DS was taken together in round 2-4, because, based on round 1, we did not expect participants' opinions about the quality domains to be different regarding children and adults.
[c] Percentage of participants that had answered "very important" (1) or "important" (2) if patient representatives are not included in analysis. The difference was only showed if exclusion of patient representatives resulted in a different conclusion regarding consensus.
[d] A medical guideline for adults with DS is at this moment being developed in the Netherlands

Appendix 4-IV

Table 4-IV Extent to which consensus was achieved among participants regarding: Health care services/disciplines

	Round 1				Round 2				Round 4
About which health care services / disciplines should the QIs provide information? *(How important do you consider the following services / disciplines for QIs?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**									
	Children		**Adults**		**Children**		**Adults**		
	% 1-2[a]	**median**	**% 1-2[a]**	**median**	**% 1-2[a]**	**median**	**% 1-2[a]**	**median**	
Downteam child	**100**	**1**			**100**	**1**	n/a	n/a	
Downteam adult			**90,6**	**1**	n/a		**85,7**	**1**	
Paediatrician	**100**	**1**			**85,7**	**1**		n/a	
ID physician			**100**	**1**	42,9	3	**96,4**	**1**	
Physiotherapist	**96,4**	**1,5**	63	2	**78,6**	**2**	50	2,5	
Speech therapist	**96,7**	**1**	70,4	2	**89,3**	**2**	60,7	2	
Dietician	**82,8**	**2**	**81,5**	**2**	64,3	2	**78,6**	**2**	
Occupational therapist	69,2	2	50	2,5	50	2,5	32,1	3	
Podiatrist	54,2	2	50	2,5	39,3	3	46,4	3	
Dermatologist	48	3	56,5	2	21,4	3	35,7	3	
Psychologist	**93,3**	**2**	**96,4**	**2**	**92,9**	**2**	**89,3**	**2**	
Youth health care (municipal health service)	46,2	3			28,6	3	n/a	n/a	
Dental hygienist	**80,8**	**2**	**80,8**	**2**					
Regular dentist (primary care)	64	2	69,2	2					
Special dentist (secondary care, specialised in special groups)	**79,2**	**2**	**76**	**2**					
Dentistry together					71,4	2	71,4	2	
General practitioner	71	2	**79,3**	**2**	53,6	2	64,3	2	
Palliative care			**76**	**2**	n/a		64,3	2	
Other, please add:									
Care for dementia or decline					n/a		**85,7**	**2**	

Table 4-IV continued

	Round 1				Round 2				Round 4
About which health care services / disciplines should the QIs provide information? *(How important do you consider the following services / disciplines for QIs?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**	**Children**		**Adults**		**Children**		**Adults**		
	% 1-2[a]	**median**	**% 1-2[a]**	**median**	**% 1-2[a]**	**median**	**% 1-2[a]**	**median**	
Cardiologist					71,4	2	46,4	3	
Rehabilitation physician					39,3	3	17,9	3	
Orthopaedist					39,3	3	42,9	3	
Physical education					25	3	25	3	
ENT specialist					60,7	2	35,7	3	
Hearing screening					**89,3**	**2**	**85,7**	**2**	
Ophthalmologist / orthoptist					67,9	2	64,3	2	
Optometrist					46,4	3	46,4	3	
Centre of expertise - vision					32,1	3	35,7	3	
Centre of expertise - sleeping					17,9	3	28,6	3	
Screening for coeliac disease					60,7	2	46,4	3	
Screening for thyroid dysfunction					64,3	2	60,7	2	
Screening for diabetes mellitus					39,3	3	60,7	2	
ID specialised nursing					25	3	42,9	3	
Support staff in assisted living facility					46,4	3	71,4	2	
Family support					**75** 73,1[b]	**2** 2	17,9	3	
Case manager					64,3	2	**85,7**	**2**	

Table 4-IV continued

	Round 1				Round 2				Round 4
About which health care services / disciplines should the QIs provide information? *(How important do you consider the following services / disciplines for QIs?)* **1=very important, 2=important, 3=neutral, 4=not that important, 5=not important at all**									
	Children		**Adults**		**Children**		**Adults**		
	% 1-2[a]	**median**	**% 1-2[a]**	**median**	**% 1-2[a]**	**median**	**% 1-2[a]**	**median**	
To what extent do you agree with the following propositions? **1=totally agree, 2=agree, 3=neutral, 4=disagree, 5=totally disagree**									
					Children and adults together[c]				
					% 1-2[a]	**median**			
QIs are different for each life phase					**89,3**	**2**			
QIs should also cover non-medical disciplines									65,4 2
QIs should include all disciplines involved in health care for people with DS					**78,6**	**2**			
Health care elements are more important for the QI-set when:									
• More people with DS need them					**96,4**	**2**			
• They contribute more to QoL					**100**	**1**			
• There are more providing professionals					32,1	3			
• There are more doubts about the quality of the element					**89,3**	**2**			

Abbreviations: QI=Quality indicator; ID=Intellectual disability; ENT=Ear nose throat; DS=Down syndrome; QoL=Quality of life.
Empty fields indicate that the topic was not presented to the participants in the concerning round.
[a] %1-2 indicates the percentage of participants that had answered "very important"/"totally agree" (1) or "important"/"agree" (2). If the percentage was ≥ 75 and the median was ≤ 2, there was consensus among the participants. Consensus was indicated in **bold**.
[b] Percentage of participants that had answered "very important" (1) or "important" (2) if patient representatives are not included in analysis. The difference was only showed if exclusion of patient representatives resulted in a different conclusion regarding consensus.
[c] For these propositions, health care for children and adults with DS was taken together, because we did not expect participants' opinions about these propositions to be different regarding children and adults.

Appendix 4-V

Table 4-V Preferred number and type of QIs and extent to which consensus was achieved among participants regarding related propositions

	Round 2	Round 3		Round 4	
How many QIs should the set contain? **Median; mean [range]** **(without patient representatives)**					
Total number	10; 6,1 [5-40] (idem)				
Number in basic set				10; 7,3 [5-30] (10; 7,9 [5-30])	
Number per module				10; 7 [3-20] (10; 7,6 [3-20])	
What type of QIs should the set contain? **Mean [range]** **(without patient representatives)**					
Structure	16,2 [5-30] (16,8)	15,8 [5-30] (idem)			
Process	19,5 [10-30] (19)	21,3 [5-35] (idem)			
Outcome	24,4 [10-40] (24,1)	22,9 [10-50] (idem)			
Distribution Structure : Process : Outcome	27%:33%:40% (28%:32%:40%)	27%:35%:38% (idem)			
		% 1-2[a]	**median**		
To what extent do you agree with the following proposition? **1=totally agree, 2=agree, 3=neutral, 4=disagree, 5=totally disagree**				**% 1-2**[a]	**median**
The QI-set should be modular in order to allow users to choose which information should be registered to serve their purpose		**81,5**	**2**		
Next to a general set containing indicators of all disciplines, there should be an elaborated set per discipline		**81,5**	**2**		
There should be a basic set of indicators, consisting of indicators that are relevant for all people with DS				**92,3**	**1,5**
Next to this basic set, additional modules should be present for specific health care or patient groups				**84,6**	**1,73**
Well defined outcome indicators are able to provide insight into process and structure too				53,8	2

Abbreviations: QI=Quality indicator; DS=Down syndrome.

Empty fields indicate that the topic was not presented to the participants in the concerning round.

[a] %1-2 indicates the percentage of participants that had answered "totally agree" (1) or "agree" (2). If the percentage was ≥ 75 and the median was ≤ 2, there was consensus among the participants. Consensus was indicated in **bold**.

Appendix 4-VI

Table 4-VI Extent to which consensus was achieved among participants regarding: Information sources and transparency of QIs and practical issues regarding development

	Round 3		Round 4	
To what extent do you agree with the following propositions? **1=totally agree, 2=agree, 3=neutral, 4=disagree, 5=totally disagree**				
	% 1-2[a]	**median**	**% 1-2[a]**	**median**
Standardisation and interoperability of electronic medical records needs to be established before quality can be measured			19,2	3
Burden for people with DS and their caregivers should be as low as possible when measuring quality			**76,9**	**2**
Burden for health care professionals should be as low as possible when measuring quality			69,2	2
People with DS (+ caregivers) and health care professionals should both deliver information for the QIs.			**100**	**2**
Parents/caregivers should themselves be responsible for documenting and keeping track of needed health care for the person with DS			**76,9**	**2**
A dialogue between health care professional and person with DS should be used as instrument for measuring customer satisfaction			73,1 **75[b]**	2 **2**
An instrument measuring patient experiences or satisfaction should be suitable to be filled out by 80% of the DS-population by themselves			46,2	3
When people with DS are not able to provide quality information themselves, their legal representative should decide who is eligible to provide this information			**92,3**	**2**
Quality information should be available on organisational/ departmental level	**77,8**	**2**		
Quality information should be available on provider level	44,4	3		
There should be a quality mark for providers / organisations specialised in DS	63	2		
Health care organisations should publish quality information on their websites	**81,5**	**2**		
QIs should only measure quality of health care provided by health care professionals with regular contact with people with DS	37	3		
Applying for joining the QI-set should be voluntary, and could be an opportunity for health care providers to display their expertise	66,7	2		
Quality information should be public on the organisational level, but not on provider (personal) level			**80,8**	**2**
Health care professionals should themselves decide about public availability of quality information			**76,9**	**2**
Privacy of professionals should be protected just as much as privacy of patients			**84,6**	**2**
QIs should stimulate improvement of care and not judge health care professionals			**92,3**	**2**
Professionals should be obliged to register the QIs if they want to be seen as 'DS-specialised'			**88,5**	**1**
Professionals wanting to be seen as 'DS-specialised' should make their quality information publicly available			69,2	2
Publishing quality information will not result in long waiting lists since most people with DS / parents will not be willing to travel far for better care.			3,8 (61,5 4-5)[c]	4

Abbreviations: QI=Quality indicator; DS=Down syndrome.
Empty fields indicate that the topic was not presented to the participants in the concerning round.
[a] %1-2 indicates the percentage of participants that had answered "totally agree" (1) or "agree" (2). If the percentage was ≥ 75 and the median was ≤ 2, there was consensus among the participants. Consensus was indicated in **bold**.
[b] Percentage of participants that had answered "totally agree" (1) or "agree" (2) if patient representatives are not included in analysis. The difference was only showed if exclusion of patient representatives resulted in a different conclusion regarding consensus.
[c] % 4-5, indicating the percentage of participants that had answered "disagree" (4) or "totally disagree" (5).


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Towards quality indicators for health care for people with Down syndrome

Submitted as:
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ABSTRACT

Objective: Health care delivery does not always fully meet complex health care needs, as is the case for people with Down syndrome (DS). In an attempt to improve this, the Dutch research project 'inDicatorS' aims to formulate quality indicators (QIs) that are relevant for people with DS, their caregivers and professionals, while providing insight into quality of health care for people with DS.

Methods: inDicatorS comprised three studies: a literature review, a qualitative exploration involving (caregivers of) people with DS, and a Delphi-study among professionals and patient organisations. We integrated the results of these studies to operationalise health care quality from the perspective of people with DS, their caregivers, and professionals, and grouped the operationalisations into three main quality dimensions: 1) effectiveness; 2) organisation of care; and 3) person-centredness. For each operationalisation, we drafted potential QIs, which were refined based on appraisal by experts in the field of health care for people with DS. Additionally, we composed a framework for future development of QI-sets.

Results: 29 operationalisations and 46 corresponding QIs were formulated. Consulted experts considered these QIs relevant and suitable for improving health care for people with DS. They suggested that limiting the number of QIs, preventing high administrative burden by facilitating easy data collection, and finding the right balance regarding transparency of quality information should be considered in the future implementation of QIs. They also noted that for some quality operationalisations, quality instruments other than QIs would be more obvious.

Conclusions: inDicatorS provides an evidence- and practice-informed basis for the further refinement and implementation of QI-sets for health care for people with DS. The proposed QIs and the framework for further development and implementation provide policy makers, health insurers, managers and professionals with directions to bring quality of health care for people with DS, and other people with complex health care needs, to a higher level.

INTRODUCTION

Despite many health care improvement initiatives, often a gap remains between health care delivery and health care needs, especially when these needs are complex (Amalberti et al., 2019; Braithwaite et al., 2019; Poitras et al., 2018; Santana et al., 2018). People with Down syndrome (DS) are a typical example. DS is the most prevalent genetic cause of intellectual disability (ID) and is related to a large variety of comorbidities, such as hearing and eye problems, thyroid dysfunction, psychological problems, heart defects, and joint problems (Capone et al., 2018; Capone et al., 2020). Consequentially, people with DS may receive health care from a large variety of health care providers, such as a paediatrician, ID physician, general practitioner (GP), physiotherapist, speech therapist, psychiatrist, ophthalmologist, and cardiologist. Additionally, they may receive health care from a DS-specialised multidisciplinary team in which health care professionals – in various compositions – provide medical advice and screening to either children or adults. In the Netherlands, such a team is referred to as 'Downteam'.

Although quality of health care for people with DS needs special attention (Grieco et al., 2015; Kinnear et al., 2018; Skotko et al., 2013), there appears to be a knowledge gap in this area (Lavigne et al., 2015; Van den Driessen Mareeuw et al., 2017). It is clear, however, that the complex health care needs of people with DS are not always sufficiently answered (Capone et al., 2018; Coppus, 2017; De Graaf et al., 2017; Grieco et al., 2015; Phelps et al., 2012). Our study aims to contribute to the knowledge on health care quality for people with DS from the perspective of patients and their families as well as from the professional perspective.

Health care quality is a multidimensional concept (Kelley & Hurst, 2006). The World Health Organization (WHO) defined six dimensions of quality of care: being effective, efficient, accessible, acceptable/patient-centred, equitable, and safe (WHO, 2006). Each dimension may include several sub-dimensions (Rawson & Moretz, 2016; Singer et al., 2011).

Quality indicators (QIs) aim to provide insight into health care quality and may identify opportunities for improvement (Boulkedid et al., 2011; Campbell et al., 2011; Donabedian, 2005). QIs can be a measure for structures, processes, or outcomes of health care, respectively referring to the setting in which health care

takes place and the available resources, the way in which health care is provided (e.g. therapeutic procedures), and the results of the provided health care (e.g. improved health) (Donabedian, 2005). Together with guidelines, QIs can serve as a framework for quality management (Boulkedid et al., 2011; Campbell et al., 2011; Donabedian, 2005).

We started the inDicatorS-project in the Netherlands to lay the groundwork for the development of QIs that contribute to health care improvements that truly matter to people with DS of all ages and life phases and with different complex needs. So far, the project comprised three studies: 1) a literature review searching for existing QIs for health care for people with DS (Van den Driessen Mareeuw et al., 2017); 2) a qualitative exploration involving interviews and focus groups with people with DS and their parents and support staff (Van den Driessen Mareeuw et al., 2020b); and 3) a Delphi study including health care professionals and patient organisations' representatives (Van den Driessen Mareeuw et al, 2020a). Our key purposes for future QIs were to 1) inform people with DS and their caregivers about available health care in order to choose the best suitable health care; 2) inform health care providers about availability and quality of DS-specialised providers to promote appropriate referrals; 3) inform teams / organisations about potential areas for internal quality improvements (Van den Driessen Mareeuw et al., 2020a&b).

The WHO dimensions of quality of care (WHO, 2006), and Donabedian's framework distinguishing structure, process and outcome measures (Donabedian, 2005) formed the theoretical framework of the project. Additionally, the project was informed by Dutch guidelines on health care for children and adults with DS. In this paper, we integrate information from the studies, resulting in 1) a longlist containing all potentially relevant QIs, and 2) a framework for future refinement and implementation of one or more shortlists of general and specific QI sets.

METHODS

The first step in the inDicatorS-project was a literature search for existing QIs, which were, however, not found (Van den Driessen Mareeuw et al., 2017). Hence, we concluded that QIs needed to be developed from scratch. Therefore, we applied a bottom-up approach in which the primary beneficiaries, people with DS, and

people close to them, their parents and support staff (of people with DS living in assisted living facilities), formed the primary source of information. For this, we used interviews and focus groups (Van den Driessen Mareeuw et al., 2020b). Finally, in order to work towards consensus on topics that QIs need to address and on potential use cases for QIs, we conducted a Delphi study including health care professionals for people with DS and patient representative organisations (Van den Driessen Mareeuw et al., 2020a). Each of these studies informed the next. For example, findings from the interviews and focus groups were presented to participants of the Delphi study. Table 5.1 provides an overview of the three studies, their methods, aims and main results.

Table 5.1 Preceding studies

Study	Literature study [a]	Interview study [b]	Delphi study [c]
Aim	Identifying existing QIs for health care for people with DS.	Exploring the perspectives of people with DS, their parents and support staff regarding quality in health care for people with DS.	Finding consensus among participants regarding QIs for health care for people with DS.
Methods	Scoping review searching for studies concerning the development and implementation of quality indicators in the field of health care for people with DS and ID.	Semi-structured interviews with people with DS and parents; focus groups with support staff (of people with DS living in assisted living facilities).	Delphi-study including professionals and patient organisation representatives.
Main results	- No published QIs for health care for people with DS were found (except for one measuring national prevalence of thyroid dysfunction). - Existing QIs concerned people with ID in general or did not specifically focus on health care (but for instance on care in assisted living facilities).	- Large variety of health problems, - Many different health care disciplines / services involved. - Important elements of health care quality: o well-coordinated health care aligned with other support and care systems; o a person-centred and holistic approach, including respect and trust; o provider–patient communication adapted to the abilities of people with DS.	- Purposes of QIs: provide insight into available health care and enable health care improvements - Large diversity of quality domains and health care disciplines should be covered. - Limited number of QIs in the set - Public quality information on the level of individual health care professionals may harm health care quality. - Measurement instruments should be suitable for people with DS (also).

Table 5.1 continued

Study	Literature study [a]	Interview study [b]	Delphi study [c]
Conclusion(s)	QIs should be patient-centred and outcome-oriented, QIs should be developed in a multidisciplinary way.	Because of the complexity of health care for people with DS, an integrated care model could be helpful in achieving health care that is person-centred and answering the specific health care needs of people with DS.	QI-set should be able to measure quality of many disciplines with only few indicators. Public availability of quality information should not be of the expense of health care quality.
Implication(s) for inDicatorS-project	QIs had to be developed from scratch.	QIs should reflect the diversity of health care disciplines and health care quality elements mentioned by the participants. Answering needs of people with DS should be key.	Selection of QIs requires careful consideration of the issues mentioned by all relevant stakeholders.

Abbreviations: QI=Quality Indicator; DS=Down syndrome; ID=Intellectual disability/ies

[a] Literature stud: aiming to identify existing QIs for health care for people with DS (Van den Driessen Mareeuw et al., 2017).

[b] 'Patient' study: semi-structured interviews and focus groups among people with DS, their parents and support staff exploring their experiences with health care and their definition of health care quality (Van den Driessen Mareeuw et al., 2020b).

[c] Professionals study: Delphi-study among health care professionals and patient organisations' representatives (Van den Driessen Mareeuw et al., 2020a).

Drafting QIs

The previous studies yielded a large number and variety of issues related to quality of health care for people with DS. These issues were clustered into groups, or sub-dimensions, based on which operationalisations of health care quality were formulated. An example of a quality issue that had emerged from the previous studies was the importance of suitable communication skills of health care professionals, which is reflected in quality sub-dimension "Communication: Whether provider adapts communication to (dis)ability of patient and builds a trust relationship". This quality sub-dimension is operationalised as "Satisfaction of patient / caregiver regarding communication and trust relationship" and "Whether health care professionals are trained in communication with people with DS/ID".

We were able to cluster the quality sub-dimensions containing the operationalisations of quality into three main quality dimensions: 1) effectiveness; 2) organisation of care; and 3) person-centredness. These dimensions correspond roughly with the WHO dimensions effectiveness, safety, accessibility and patient-centredness, albeit that we divided patient-centredness into organisational characteristics (namely organisation, coordination and continuity) and

characteristics of the patient-provider interaction (namely person-centredness and communication).
Then, for each quality operationalisation, one or more potential QI(s) were formulated, informed by literature on (development of) indicators (Coker et al., 2012; Corrigan et al., 2001; Engels et al., 2005; Kelley & Hurst, 2006; Kötter et al., 2012; Rubin et al., 2001; Seow et al., 2009; Uddin et al., 2015).
More than one potential QI per operationalisation was formulated if information could be obtained from different information sources or at different organisational levels, or if objective (e.g. blood test results) and subjective (e.g. experienced health improvements) information could be gathered.
The QIs were then sorted by information source (people with DS plus caregivers, professionals, or other), to provide insight into how and where implementation of the QIs could take place and what kind of measurement instrument could be used. For example, the previous studies suggested that data collection from people with DS and/or their caregivers could be done via (digitalised) surveys or questionnaires. By presenting the QIs per information source, it becomes clear what kind of questions can be presented in such a survey or questionnaire. The same holds for health care professionals, who are used to record information in electronic medical record systems (EMR)s. The QIs formulated under 'professionals' reflect the items that ideally should be built into their registration systems.

In this paper, we give an overview of operationalisations based on all quality issues that emerged in the inDicatorS-project as ideally being part of a QI-set for health care for people with DS, regardless of any practical obstacles. This includes operationalisations based on quality issues mentioned by people with DS or their caregivers, by health care professionals and operationalisations based on quality issues mentioned by both. As a consequence of the diverse group of people with DS and the diversity of (health) care disciplines involved, some operationalisations may be relevant to (almost) all people with DS and all (health) care disciplines involved, while others may only be relevant to a specific group. In order to address this, an overview was made indicating which disciplines are indispensable to (almost) all people with DS and which are only needed by a selection of people with DS having specific health problems. We did not include suggestions made by the study participants about general legal regulations, such as whether a professional is registered in the national professional register, or the presence of a complaints procedure, and discipline-specific operationalisations (e.g. procedure

for ophthalmological examinations or heart surgery), because these topics are covered by national or discipline-specific audits, indicators or guidelines.

A framework for further refinement and implementation

Next to the operationalisations of quality to be reflected in future QIs, the three preceding studies provided insight into considerations and circumstances that should be taken into account during further refinement and implementation of QIs in health care for people with DS in the Netherlands, but potentially in other health care systems as well. These considerations and circumstances were combined to form a framework for future QI development.

Practical appraisal

In order to obtain insight into how the potential QIs are appraised in practice, the list of sorted QIs (per information source) was presented to representatives of relevant professional and patient organisations (n=22). These representatives, having expertise in health care for people with DS, had been involved throughout the inDicatorS-project as critical reviewers and had been recruited through and by their professional / patient organisation. The representatives were asked to comment on the list of potential QIs as a whole, in terms of feasibility (whether they thought the QIs could be implemented/used in practice), and in terms of desirability (whether they thought the QIs could improve the quality of health care for people with DS). Representatives could also comment on specific QIs. Furthermore, representatives were asked to assess whether they thought the QIs would be able to contribute to the main purposes of QIs: providing (caregivers of) people with DS and professionals with information on available health care, and providing information that could be used for improvements in health care provision.

Appendix 5-I shows the list of sorted potential QIs that was presented to the representatives. The comments of the representatives were compared and clustered per theme, resulting in suggestions for refinements of the QIs and for implementation and use. Based on these suggestions, some refinements were applied to the potential QIs and to the framework for further development.

RESULTS

Practical appraisal

Of the twenty-two representatives who participated in the practical appraisal, eighteen thought that the QIs would be able to contribute to generally better health care for people with DS, twenty thought that the QIs would be useful for improvement initiatives for, and by, professionals, and thirteen thought that the QIs would be feasible.

In their comments, representatives explained that all relevant quality dimensions regarding health care for people with DS were covered by the QIs. Furthermore, they liked the fact that the listed QIs involved data collection from both patients and professionals (and other sources/registries), as this would provide a complete and balanced insight into quality. However, several representatives thought that the large number of QIs and the fact that they were sorted per information source, hampered conciseness. Other remarks were that the QIs were too general and too little DS-specialised, that much work is still to be done in order to implement and use the QIs, and that information on how the QIs should be put into practice was lacking. For example, representatives noted that a QI measuring whether professionals completed DS-specialised education would be redundant, if no such education is present. Likewise, although the proposed QIs contain measures on the regional level (e.g. the number of DS-specialised health care within one region), such QIs would not be useful if there are no registries for regional DS-data. Nevertheless, regional collaboration initiatives, in which necessary disciplines/experts can be easily involved or consulted, were much welcomed and encouraged by the representatives. Additionally, for some of the operationalisations of quality it was suggested not to develop QIs. For example, according to the representatives, coordination and organisation of care could better be addressed through structured discussion in evaluation meetings of health care professionals, not necessarily in a measurement instrument. Other quality issues could be addressed by means of a dialogue between a health care professional and the person with DS (and their caregivers). Furthermore, it was mentioned that instead of, or additional to, QIs, the identified quality operationalisations could be incorporated in a checklist that people with DS or caregivers could use to evaluate the received care and as input for the dialogue with health care professionals.

The representatives also suggested to add and/or refine some QIs. Appendix 5-II shows these suggestions and the refinements that were applied based on these

suggestions. For example, among other things, the classification according to information source was deleted in order to increase conciseness and several QIs were reformulated.

Operationalisation of quality and potential QIs

Table 5.2 shows the 29 operationalisations of quality that were formulated based on the findings from the previous studies and the practical appraisal of potential QIs, grouped by the three main quality dimensions and their sub-dimensions, as described above. A total of 46 potential QIs were drafted, which are shown in Appendix 5-III. The table in Appendix 5-III also shows the organisational level(s) to which potential QIs could apply (i.e. health care professional, health care organisation/practice, Downteam, community/region/national), as well as potential data sources or instruments for collection of data for the QIs (e.g. patient survey, EMR, other databases/registries). Additionally, the table indicates the type (structure, process, outcome) of each QI. This shows that most QIs are measures for structure or process and only a few measure outcomes. Finally, the table in appendix 5-I shows which QIs are relevant only for people with DS and which could also be relevant for people with complex diseases in general. QIs specifically relevant to people with DS address: adherence to national DS-guidelines (QI: 1.1c), DS-specific expertise of professionals (QIs: 1.2a, 1.2b-I), Downteams (QIs: 2.2a, 2.2b, 2.2c); presence of a DS-specific transition protocol for the transition from paediatric to adult care (QI: 2.3d-I&II)); the number of DS-specialised health care professionals in the region 2.4a-I). QIs relevant for people with DS as well as other people with complex disease address: presence of a case manager (QIs: 2.1b, 2.1c-I&II); Transition from paediatric to adult care (QIs: 2.3a, 2.3b, 2.3c); need/availability of family support (QIs: 3.2b-I&II). The remaining QIs may also be relevant for patients in general.

Table 5.2 Quality operationalisations to be measured by quality indicators, grouped per quality dimension and quality sub-dimension

1	**Effectiveness**
1.1	*Effectiveness - Timely recognition of health problems: Whether health problems are sufficiently and timely recognised and treated* a. Satisfaction of people with DS / their caregivers about health / quality of life b. Objective health outcomes (e.g. outcomes of blood test, physical examination) c. Adherence to guidelines (e.g. are recommended screenings performed and frequency of contact moments with specific professionals) or reasoned deviation d. Time between signalling of health problem and treatment
1.2	*Effectiveness - Expertise of providers* a. DS-specific training / education completed by health care professionals and/or their professional experience with people with DS. b. Expertise experienced by people with DS / their caregivers
1.3	*Safety* a. Availability of safety protocols
2	**Organisation of care**
2.1	*Organisation, coordination and continuity in general* a. (multidisciplinary) collaboration or coordination networks b. Presence of an internal case manager c. Presence of a (regional) case manager d. The presence of a care plan e. Whether referrals are made easily f. Experienced (multidisciplinary) collaboration, coordination, and continuity within the organisation
2.2	*Organisation, coordination and continuity specifically for Downteams* a. Distribution of multidisciplinary composed Downteams b. Collaboration or coordination c. Case manager
2.3	*Organisation, coordination and continuity specifically related to transition from child to adult care* a. Whether data are transferred from paediatrician to ID physician / general practitioner b. Whether patients are satisfied about transition in health care c. Whether patients are satisfied about transition in non-health care d. Presence of a transition protocol
2.4	*Accessibility – Health care nearby / within community or in primary care centres* a. Distribution of DS-specialised health care providers per region b. Presence of a usual source of care
3	**Person-centredness**
3.1	*General* a. Whether health care professionals are trained in person-centredness
3.2	*Impact/burden of health care/treatment on patient's life and on his/her environment* a. Whether provider maps the personal situation and adapts treatment/advice/support accordingly b. Health care providers make sure family support is being offered if needed.
3.3	*Involvement of all relevant stakeholders* a. Whether providers involve patients/caregivers/other providers in decisions
3.4	*Consideration of preferences and values of the person with DS and his/her family* a. Whether person with DS / caregivers feel their values/preferences/worries are taken into account.
3.5	*Communication: Whether provider adapts communication to (dis)ability of patient and builds a trust relationship* a. Satisfaction of patient / caregiver regarding communication and trust relationship b. Whether health care professionals are trained in communication with people with DS/ID

Applicable to both children and adults if not specified otherwise
Abbreviations: DS=Down syndrome; ID=Intellectual disability/ies.

A framework for future refinement and implementation of QI-sets

The proposed QIs form a 'longlist' containing all elements needed to provide a complete picture of quality of health care for people with DS. There are, however, practical constraints and considerations related to the use of these QIs, which emerged from our studies and the practical appraisal of potential QIs. We took these into consideration when forming a framework for the translation of operationalisations of quality into QIs and for the future implementation of developed QI-sets.

From the previous studies, it became clear that not only professionals should be actively involved in the actual development and selection of QIs, but also people with DS and their caregivers, as well as researchers and health insurers. Health care professionals –particularly those with a core function in health care for people with DS- could take the lead in a joint effort of all relevant stakeholders to select, develop and implement QIs. Table 5.3 shows the health care disciplines which were considered by the participants in our studies as crucial for all people with DS (having a core function in health care for people with DS) and the disciplines which would only be crucial for some people with DS.

Table 5.3 Health care disciplines considered to be indispensable to all people with DS and to certain subgroups according to the study participants

Health care discipline considered to be indispensable for	all children with DS	all adults with DS	children with DS with special health care needs only	adults with DS with special health care needs only
Downteam	x	x		
Paediatrician	x			
ID physician		x	x	
Physiotherapist	x			x
Speech therapist	x			x
Dietician	x	x		
Psychologist	x	x		
Dentist	x	x		
General practitioner	x	x		
Audiologist	x	x		
Palliative care		x	x	
Care for decline / dementia		x		
Family support	x			
Case manager		x		
Occupational therapist			x	x
Podiatrist			x	x
Dermatologist				x
Cardiologist			x	x

Table 5.3 continued

Health care discipline considered to be indispensable for	all children with DS	all adults with DS	children with DS with special health care needs only	adults with DS with special health care needs only
Rehabilitation physician			x	x
Orthopaedist			x	x
Physical education			x	
ENT specialist				x
Ophthalmologist / Optometrist / Orthoptist			x	x
Centre of expertise – vision			x	x
Centre of expertise – sleeping			x	x
Nurse specialised in intellectual disability				x
Neurologist			x	x
Urologist			x	x

Limited number of QIs

The most important constraint is the large number of identified operationalisations of quality. Developing, implementing and using QIs on all of the identified elements would be challenging (Corrigan et al., 2001; Kelley & Hurst, 2006). It would lead to a high administrative burden for professionals and (caregivers) of people with DS and would also require the development of (too many) instruments for the collection of the necessary data.

Limit administrative burden experienced by professionals

The administrative burden of using QIs may be limited by allowing professionals or practices/hospitals to make a selection of the QIs they want to use (i.e. quality issues on which they want to collect information for improvements). They may also decide to use a selection of QIs for a certain period of time and after that switch to another selection of QIs. However, the practical appraisal participants warned that this may lead to low comparability between health care professionals/practices/hospitals. Moreover, some quality elements, such as elements especially relevant to people with DS, may become underexposed. It was therefore suggested to define, in collaboration with professionals, (caregivers of) people with DS and health insurers, a core set of QIs that should at least be measured by all users. This could be facilitated by grouping QIs into modules. Such modules could, for example, include QIs relevant to all people with DS, and specific modules including QIs on health care only relevant to specific groups, such as people with DS in a certain age group or with specific health problems. Additionally, modules could be composed around specific dimensions, such as communication, or coordination.

Our previous studies indicated that, in order to limit administrative burden of professionals and stimulate accurate registration, QIs should use information that can be easily registered (e.g. by checking only a few boxes) or reuse information that is already being registered in EMRs. Additionally, registered data from different sources (e.g. from different disciplines, or other sources, such as general registries or databases and people with DS and their caregivers) should be integrated. This could be facilitated by automated extraction of data from EMRs (Borusiak et al., 2019). However, despite development of advanced data techniques, it is still difficult to (automatically) extract useful information from EMRs, which may be partly caused by inaccurate registration by health care professionals (Verheij et al., 2018). Furthermore, in the Netherlands, different disciplines and health care organisations use different EMRs that are generally not interoperable. This may be problematic given the different disciplines involved in DS.

Limit administrational burden experienced by people with DS and their caregivers

Mentioned examples of instruments for collecting the needed information from people with DS are Patient Reported Experience Measures (PREMs) and Patient Reported Outcome Measures (PROMs) (Breckenridge et al., 2015). Such instruments should be as concise and easy as possible in order to limit administrative burden of (caregivers of) people with DS.

Study participants furthermore preferred instruments that are suitable to obtain information from people with DS themselves. If this is not possible, the legal representative should provide the information, or decide who is entitled to do so. Collecting information from both people with DS and caregivers may also be an option. Participants however wondered how to handle differences between information from people with DS and caregivers. Santoro et al. (2021) for example found that adults with DS self-report a higher global health score than the score reported by their parents (Santoro et al., 2021).

It was also suggested to use existing instruments for people with ID in order to facilitate the collection of information from people with DS and their caregivers, and the development of instruments for that. Participants of our studies also mentioned that this would ascertain that the QIs align with related initiatives for people with ID, which they considered preferable. They also underlined that QIs should match existing guidelines for health care for people with DS.

Presentation of information

There was an important concern among participants of our studies related to transparent presentation of quality information obtained through QIs. Professionals feared that publicly available quality information, especially on the level of individual health care professionals, would shift market shares, potentially leading to waiting lists for high scoring providers, and hampering quality improvement. However, it was also acknowledged that professionals who want to brand themselves as 'DS-specialised', should disclose core quality information. Additionally, it was noted that for quality improvement comparison between health care professionals or organisations through sharing of information is helpful. Other professionals commented that as an alternative for full transparency and public disclosure, professionals may only reveal that they collect information for the QIs, indicating a certain awareness of specific quality elements. Nevertheless, it should be carefully determined which quality information, on what level, and to what extent should be made public in order to serve the identified purposes of the QIs.

DISCUSSION

The inDicatorS-project lays the groundwork for the development of QIs that contribute to health care improvements that truly matter to people with DS of all ages and life phases. In this paper, we described the final step of the inDicatorS-project, in which results of the previous studies are combined and reflected upon by stakeholders. This resulted in the operationalisation of quality in health care for people with DS, a longlist of potential QIs, and in a framework for further development and implementation of QIs. 29 operationalisations of quality were identified and a total of 46 QIs were formulated, which were categorised into the three quality dimensions: 1) effectiveness; 2) organisation of care; and 3) person-centredness (see Appendix 5-III). The framework provides considerations to take into account when selecting, further developing and implementing QIs: limiting the number of QIs, preventing high administrative burden by facilitating easy data collection, and finding the right balance in transparency of quality information.

Strengths and limitations

We were the first to propose development of QIs for health care for people with DS (Van den Driessen Mareeuw et al., 2017). To the best of our knowledge, no other initiatives are addressing this topic. Since we base the QIs on the results of three studies thoroughly exploring the perspectives of people with DS and their caregivers as well as their health care professionals, the QIs are meaningful and useful for both groups. Engaging health care professionals as well as patients and their families is an important prerequisite for successfully developing quality instruments in health care (Delnoij et al., 2010; Wiering et al., 2017). Furthermore, the fact that the QIs are linked to existing DS-guidelines, to which stakeholders are committed, supports the future use of these QIs in the Netherlands.

The status of the QIs (proposed QIs, not a 'ready-to-use' QI-set) can be seen as a limitation, because subsequent actions are needed before the QIs can be implemented and used. However, it can also be seen as a strength, because it leaves ample room to adapt them to the practical context(s). Literature recommends that QIs should be based on scientific knowledge and guidelines (Kötter et al., 2012; Mainz, 2003), should fit in the social, cultural and regulatory context (Engels et al., 2005), and should be representative to the entire range of health problems, health care disciplines involved, steps in the care process, and the entire life span (Corrigan et al., 2001; Kelley & Hurst, 2006; Seow et al., 2009). The thorough exploration as part of the development of our proposed QIs (previous studies) already largely covers these recommendations. The preliminary status of our proposed QIs enables even further adaptations that may result from the consultation and authorisation process following this project. Moreover, the preliminary status of the QIs can be well-explained by the relatively unexplored status of health care for people with DS (Van den Driessen Mareeuw et al., 2017; Santoro et al., 2021), which required development of QIs that started from scratch. Hence, important steps in developing QIs, such as determining scope, users, and purposes of the QIs (Kötter et al., 2012; Rubin et al., 2001), identifying relevant quality dimensions to be measured (Kötter et al., 2012), and identifying barriers and facilitators for implementation and use (Berwick, 2016; Kelley & Hurst, 2006; Seow et al., 2009), had to be, and were, carried out thoroughly.

Reflection on results

The proposed QIs are based on what people with DS, their caregivers, health care professionals, and patient organisation representatives, consider key elements of

quality in health care for people with DS. These elements or quality dimensions resemble quality dimensions defined in the literature (Kelley & Hurst, 2006; Rawson & Moretz, 2016; Singer et al., 2011; WHO, 2006). However, not all dimensions mentioned in the literature receive equal attention in our QIs. The number of QIs dedicated to 'person-centredness' is relatively large. This is in line with the current developments in health care quality, in which person- or patient-centredness is increasingly considered as a crucial element (Berwick, 2016; Santana et al., 2018). Despite this, person-centred QIs are scarce (Santana et al., 2018). In that sense, our proposed QIs are innovative, and may serve as an example for person-centred QIs, also for health care for other groups than people with DS. This is supported by the suggestion to use quality of care for the DS population as an indicator for the quality of a health care system in general (Phelps et al., 2012). The number of QIs dedicated to the dimension 'organisation of care' is also relatively large. This reflects the complexity of health care for people with DS (Minnes & Steiner, 2009), and related problems experienced by people with DS, their caregivers, and health care professionals.

The QIs are formulated for different organisational levels: individual (health care professional) level, the level of providers (hospital (departments); practices), and regional or national level. This matches the current literature on health care quality in which QIs are part of a multi-level 'learning health care system' (Menear et al., 2019) (or 'knowledge ecosystem' (Elliot et al., 2014), or 'evidence ecosystem' (Lewin et al., 2018).) Such a multi-level, systemic approach for describing health care (Menear et al., 2019), also fits our results with respect to the multidisciplinary nature of the proposed QIs, including QIs measuring (collaboration with) non-health care elements, such as social care. This reflects the expressed need for an integrated care system in which the different types of care are coordinated around people's needs. This is considered especially important for people with complex needs (González-Ortiz et al., 2018), such as people with DS (Coppus, 2017; Phelps et al., 2012).

In learning health care systems, summaries of performance against evidence-based standards are used in audits and feedback (Shepherd, 2014). Learning health care systems are seen as a pathway towards value-based health care (Menear et al., 2019). Porter (2010) defines value as health outcomes per dollar spent and outcomes are defined as the result of health care structures and processes (Donabedian, 2005). However, outcome measures do not indicate the origin of

the measured outcome and a EU Expert Panel recently acknowledged that - apart from health outcomes - the process of care may be very relevant to patients too (EXPH, 2019; Porter, 2010), especially to patients with complex needs (RVS, 2020). Another study also showed that for patients both processes and structures of care are important (Rademakers et al., 2011). This is corroborated by the relatively large number of process and structure QIs that resulted from our study.

Next steps

The proposed QIs form a firm basis for future improvement of health care for people with DS. The next step is to prioritise the proposed QIs for further development and use. For this, all stakeholders involved in health care for people with DS are needed: people with DS, their caregivers, (health care) professionals, managers in health care, health insurers, the inspectorate, and other national, regional and/or local stakeholders. Joint effort by all stakeholders is required to obtain (financial) means for integrating the QIs in EMRs and developing measurement instruments, such as PREMs or PROMs or others.

Based on the findings of our study, three priority actions can be identified. The first is to integrate the proposed QIs into existing EMRs, since part of the needed information is currently already available in EMRs, and needed technologies are present (Borusiak et al., 2019). The second is to identify and delineate existing instruments that might be suitable for collecting information from people with DS and/or their caregivers. Examples are an online tool for caregivers of people with DS (Majewski et al., 2021) and the use of global health measures in DS clinics (Santoro et al., 2021). The third is to further explore quality issues that are, according to our findings, in need of innovation, such as regional expertise networks and DS-specialised education. Such expertise related innovations may benefit from sharing information with (inter)national networks such as the European Reference Network for Intellectual disability, Telehealth, Autism and Congenital Anomalies (ITHAKA, 2021).

However, next steps in the development of the proposed QIs are also largely depending on the current societal and health care situation. For example, current anti-registration movements (Berwick, 2016; Ploegman et al., 2019) may discourage the introduction of additional QIs. Obviously, the most striking current condition in health care is COVID-19 (Auener et al., 2020; Hüls et al., 2021). For people with DS and their caregivers, this has had a large impact, because people with DS are at higher risk for (severe) medical complications if infected

by the virus (Hüls et al., 2021) and because of their difficulties deploying coping mechanisms for changing circumstances (Patel et al., 2018). More generally, the restrictions related to COVID-19 impacted whether and how acute and elective health care proceeded (Auener et al., 2020). The COVID-pandemic has put the health care system under a microscope and brought prevalent underlying ideas to the surface. For people with DS, it showed the crucial importance of well-organised care. Health care providers were forced to provide the most necessary care only and to use telemedicine or other techniques. Also, the importance of collaboration and sharing information became clear. For the further development and implementation of our proposed QIs this may imply a shift from attention to quality issues related to medical care towards quality issues related to coordination, collaboration and sharing information. Literature suggest that the COVID-pandemic has created the right moment for change (Auener et al., 2020; Dawson et al., 2021; Subbian et al., 2021). Hence, this may be the right moment to put into practice the various opportunities for change in health care for people with DS, as unveiled by our study.

Conclusion

We operationalised quality of care for people with DS and proposed QIs that are evidence-based as well as practice-based. This study forms a firm basis for future development of QI-sets, their implementation and actual use, while also showing potential shortcomings of QIs and considerations to take into account. The study provides directions to bring quality of health care for people with DS to a higher level, while fostering properly answered health care needs. This will ultimately contribute to a better life for people with DS, and potentially also for other patient groups with complex needs.

APPENDICES

Appendix 5-I

QIs sorted per information source, as presented to participants in the practical appraisal phase of the inDicatorS-project

A. Information source: people with DS, their parents or representatives

Quality dimension: **Effectiveness, accessibility and safety**	
Indicator(s):	***Numerator / denominator***
Sub-dimension: *Timely recognition of health problems: Whether health problems are sufficiently and timely recognised and treated*	
Satisfaction of people with DS or their caregivers about health or quality of life	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that is satisfied about the improvement of their health or quality of life after treatment.
Time between onset of health problem and treatment	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, with a specific health problem, whose health problem is treated within reasonable time, or who is referred to the right health care professional in time.
Sub-dimension: *Expertise of providers*	
Expertise experienced by people with DS / their caregivers	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that is satisfied about the expertise of the health care professional(s) they last visited (in one health care organisation or practice)
Sub-dimension: *Health care nearby / within community or in primary care centres*	
Distribution of DS-specialised health care providers per region	The number of patients with DS within one region that is satisfied about the proximity of needed health care professionals.
Presence of a usual source of care	Number of people with DS within one region / nationally having a place where he/she usually goes when ill / in need of advice
Sub-dimension: *Safety*	
-	
Quality dimension: **Organisation, coordination and continuity**	
Indicator(s):	***Numerator / denominator***
Sub-dimension: *Transition from child to adult care*	
Whether data are transferred from paediatrician to ID physician / GP	The number of patients with DS aged 16-23 years that went out of paediatrician's care having a record of data transference to ID physician or other health care professional.
Whether patients are satisfied about transition	- The number of patients with DS aged 16-23 years that is satisfied about the transition in health care. - The number of patients with DS aged 16-23 years that is satisfied about the transition in non-health care.
Sub-dimension: *Organisation, coordination and continuity of Downteams*	
Experienced coordination / continuity within Downteam	Number of patients with DS seen in one Downteam that is satisfied about coordination and continuity in the Downteam.
Sub-dimension: *Organisation, coordination and continuity within one health care organisation*	

Experienced coordination / continuity within the organisation	Number of patients with DS in one health care organisation that is satisfied about coordination and continuity in the organisation
The presence of a care plan	Number of patients with DS in one health care organisation having a plan, based on needs of the patient, indicating steps and time planning.
Presence of an internal case manager	Number of patients with DS in one health care organisation having a case manager who has an overview of different health care appointments
Sub-dimension: *Organisation, coordination and continuity of health care and other (non-health care) services in one region / community*	
Whether referrals are made easily	Number of patients with DS in one health care organisation that is satisfied about the convenience of referrals to other health care organisations/professionals.
Presence of a (regional) case manager	- Availability of an appointed case manager in one region who is under contract of one or more of cooperating organisations, having an overview of health care and other services helping patients finding the needed health care and services. - Number of patients with DS within that region having a case manager who has an overview of different (health)care appointments and helping patients planning their appointments.
The presence of a care plan	Number of patients with DS in one health care organisation having a plan, based on needs of the patient, indicating steps and time planning of treatment by different health care professionals
Whether patients are satisfied about coordination and collaboration	Number of patients with DS in one health care organisation that is satisfied about the convenience of referrals to other health care organisations/professionals or to non-health care services.
Quality dimension: **Person-centredness and communication**	
Indicator(s):	***Numerator / denominator***
Sub-dimension: *General*	
-	
Sub-dimension: *Impact/burden of health care/treatment on patient's life and on his/her environment*	
Whether provider maps personal situation and adapts treatment accordingly	Number of patients with DS in that organisation who feel that their situation is taken into account in deciding about treatment.
Health care providers make sure family support / early intervention is being offered if needed.	Number of patients with DS in that organisation that report that family support was discussed during the last consultation.
Satisfaction of people with DS / their caregivers about health / quality of life / participation	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that is satisfied about the improvement of their health, quality of life, or participation after treatment.
Sub-dimension: *Involvement of all relevant stakeholders*	
Whether providers involve patients/carers/other providers in decisions	Number of patients with DS in that organisation who feel that all relevant people for the decision at stake are involved.
Sub-dimension: *Consideration of preferences and values of the person with DS and his/her family*	
Whether person with DS / caregivers feel their values/ preferences are taken into account.	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that is satisfied about the extent to which their values/preferences are taken into account.
Sub-dimension: *Communication: Whether provider adapts communication to (dis)ability of patient and build a trust relationship*	

Satisfaction of patient / caregiver regarding communication and trust relationship	- Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that is satisfied about communication during last consultation. - Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that understands information provided during consultation. - Number of patients with DS (or their caregivers) experiencing a trust relationship with the health care professional most visited.

B. Information source: health care professionals, preferably via EMR[a], or professional survey.

Quality dimension: **Effectiveness, accessibility and safety**	
Indicator(s):	***Numerator / denominator***
Sub-dimension: *Timely recognition of health problems: Whether health problems are sufficiently and timely recognised and treated*	
Objective health outcomes (e.g. outcomes of blood test, physical examination)	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, that has improved objective health outcomes.
Adherence to guidelines (e.g. are recommended screenings performed and frequency of contact moments with specific professionals)	Extent to which a health care professional or health care organisation or practice adheres to guidelines. • - Screenings to be performed[b] - Periodic appointments to take place[c]
Time between onset of health problem and treatment	Number of patients with DS, seen by one health care professional or in one health care organisation or practice, with a specific health problem, whose health problem is treated within reasonable time, or who is referred to the right health care professional in time.
Sub-dimension: *Expertise of providers*	
-	
Sub-dimension: *Health care nearby / within community or in primary care centres*	
-	
Sub-dimension: *Safety*	
-	

Quality dimension: **Organisation, coordination and continuity**	
Indicator(s):	***Numerator / denominator***
Sub-dimension: *Transition from child to adult care*	
Whether data are transferred from paediatrician to ID physician / GP	The number of patients with DS aged 16-23 years that went out of paediatrician's care having a record of data transference to ID physician or other health care professional.
Sub-dimension: *Organisation, coordination and continuity of Downteams*	
Whether a multidisciplinary Downteam is present in the organisation	Presence of a multidisciplinary Downteam in the organisation
Experienced coordination / continuity within Downteam	Number of health care professionals participating in one Downteam that is satisfied about coordination and continuity in the Downteam.
Sub-dimension: *Organisation, coordination and continuity within one health care organisation*	
Experienced coordination / continuity within the organisation	Number of health care professionals in one health care organisation that is satisfied about coordination and continuity in the organisation

Sub-dimension: *Organisation, coordination and continuity of health care and other (non-health care) services in one region / community*	
Whether referrals are made easily	Number of (health care) professionals in one health care organisation or region or nationally that is satisfied about the convenience of referrals to other (health care) organisations/professionals or services.
Quality dimension: **Person-centredness and communication**	
Indicator(s):	***Numerator / denominator***
Sub-dimension: *General*	
-	
Sub-dimension: *Impact/burden of health care/treatment on patient's life and on his/her environment*	
Whether provider maps personal situation and adapts treatment accordingly	Number of health care professionals within one organisation who made a record of having mapped the personal situation and adapted treatment accordingly.
Health care providers make sure family support / early intervention is being offered if needed.	Number of health care professionals within one organisation who made a record of having checked the need for family support.
Quality of life (QoL) of the person with DS	- Number of patients with DS of one organisation whose QoL has improved after treatment in that organisation. - Number of patients with DS in a region / in the Netherlands whose QoL has improved over time.
Participation in society	- Number of patients with DS of one organisation whose participation in society has improved after treatment in that organisation. - Number of patients with DS in a region or country whose participation in society has improved over time.
Sub-dimension: *Involvement of all relevant stakeholders*	
Whether providers involve patients/carers/other providers in decisions	Number of health care professionals within one organisation who made a record of having involved all relevant people for the decision at stake.
Sub-dimension: *Consideration of preferences and values of the person with DS and his/her family*	
Whether providers take into account values/preferences of the person with DS / carers.	Number of health care professionals within one organisation who made a record of having taken into account values/preferences of person with DS.
Sub-dimension: *Communication: Whether provider adapts communication to (dis)ability of patient and build a trust relationship*	
-	

a EMR = electronic medical record, bHeart defects / functioning, Thyroid function (children), Coeliac disease (children), Hearing / ear problems, Vision / eye disorders, Dental (problems), Dementia (older adults).
c Paediatrician (children), ID physician (adults, children: if needed), Downteam child (children), Downteam adult (adults), ear-nose-throat (ENT)-physician, Ophthalmologist / Orthoptist, Dentist, Physiotherapist, Speech therapist, Dietician, Youth care (children).

C. Information source: other

Quality dimension: **Effectiveness, accessibility and safety**		
Indicator(s):	***Numerator / denominator***	Proposed source
Sub-dimension: *Timely recognition of health problems: Whether health problems are sufficiently and timely recognised and treated*		
-		
Sub-dimension: *Expertise of providers*		
Training / education completed by health care professionals	- Number of health care professionals within one health care organisation / practice having completed DS-specialised training / education. - Whether a health care professional completed DS-specialised training / education.	HRM[d]-data of health care organisation / registry
Sub-dimension: *Health care nearby / within community or in primary care centres*		
Distribution of DS-specialised health care providers per region	- The number of health care professionals participating in a QI-set per region. - The number of patients with DS within one region having needed health care professionals within reasonable distance from their homes. - The number of patients with DS within one region that is satisfied about the proximity of needed health care professionals.	National database
Presence of a usual source of care	Number of people with DS within one region or country having a place where he/she usually goes when ill or in need of advice.	
Sub-dimension: *Safety*		
Availability of and adherence to safety protocols	Whether a safety protocol is present in a health care organisation / practice providing health care to people with DS, and whether it is adhered to.	Database/registry of health care organisation and/or inspectorate/audit
Quality dimension: **Organisation, coordination and continuity**		
Indicator(s):	***Numerator / denominator***	Proposed source
Sub-dimension: *Transition from child to adult care*		
Presence of a transition protocol	- Whether the health care organisation providing paediatric care to children with DS has a transition protocol - Whether the health care organisation providing adult care to adults with DS has a transition protocol	Database/registry of health care organisation
Sub-dimension: *Organisation coordination and continuity of Downteams*		
Whether a multidisciplinary Downteam is present in the hospital /other health care organisation	Presence of a multidisciplinary Downteam in the organisation	Database/registry of health care organisation
Existence of collaboration agreements	Existence of collaboration agreements between departments or professionals participating in the Downteam	Database/registry of health care organisation
Presence of a case manager / coordinator	Availability of a case manager / coordinator of the Downteam, planning the appointments, gathering information from other health care professionals (outside the Downteam), and helping patients preparing the appointments.	Database/registry of health care organisation

Sub-dimension: *Organisation coordination and continuity within one health care organisation*		
The presence of a care plan	Number of patients with DS in one health care organisation having a care plan, based on needs of the patient, indicating steps and time planning.	inspectorate registry/ audit
Presence of an internal case manager	Availability of an appointed case manager within the organisation helping patients planning their appointments.	Database/registry of health care organisation
Sub-dimension: *Organisation, coordination and continuity of health care and other (non-health care) services in one region / community*		
Whether referrals are made easily	Number of (health care) professionals in one health care organisation or region or country that is satisfied about the convenience of referrals to other (health care) organisations/professionals or services.	Database/registry of health care organisations/services
Existence of collaboration agreements	Existence of collaboration agreements between organisations or professionals of health care and other services within one region or country.	Database/registry of health care organisation, inspectorate registry/ audit
Presence of a (regional) case manager	- Availability of an appointed case manager in one region who is under contract of one or more of cooperating organisations, having an overview of health care and other services helping patients finding the needed health care and services. - Number of patients with DS within that region having a case manager who has an overview of different (health)care appointments and helping patients planning their appointments.	Database/registry of health care organisation(s) / service(s) / regional registration
The presence of a care plan	Number of patients with DS in one health care organisation or region or country having a plan, based on needs of the patient, indicating steps and time planning of treatment by different health care professionals and needed services (outside health care).	inspectorate registry/ audit
Experienced coordination / continuity within the region / community	- Number of patients with DS in one organisation, region, country that is satisfied about the coordination and collaboration of all needed health care and services. - Number of health care professionals in one health care organisation / region / country that is satisfied about coordination and continuity in the region.	Database/registry of health care organisations/services
Quality dimension: **Person-centredness and communication**		
Indicator(s):	***Numerator / denominator***	Proposed source
Sub-dimension: *General*		
Whether health care professionals are trained in person-centredness	Number of health care professionals within one organisation who completed a training in providing person-centred care, or who obtained their person-centred care skills in another way.	Database/registry of health care organisations/services

Sub-dimension: *Impact/burden of health care/treatment on patient's life and on his/her environment*		
Availability of an instrument/ tool enabling health care professionals to map the personal situation.	Whether a health care organisation has such a checklist or other supporting instruments.	Database/registry of health care organisation
Sub-dimension: *Involvement of all relevant stakeholders*		
-		
Sub-dimension: *Consideration of preferences and values of the person with DS and his/her family*		
-		
Sub-dimension: *Communication: Whether provider adapts communication to (dis)ability of patient and build a trust relationship*		
Whether health care professionals are trained in communication with people with DS/ID	Number of health care professionals within one organisation who completed a training in communication with people with DS/ID, or who obtained their communication skills in another way.	Database/registry of health care organisation

[d]HRM = human resource management.

Appendix 5-II

Comments and proposed, and applied, amendments in the practical appraisal phase

Comments	Refinements applied
General	
The list of QIs is too long and not concise enough. Sorting per information source does not contribute to conciseness.	No sorting per information source Similar QIs formulated for different organisational levels (e.g. coordination on the level of a Downteam and on the regional level) were merged into one QI Sub-dimension 'Accessibility' was moved to 'organisation, coordination and continuity'.
Perhaps: sort by 'for children' and 'for adults'	No change, because there is too much variation among children and among adults.
Please prioritise: which QIs are necessary for all, which only for a few people with DS?	No change, we consider all QIs relevant to all people with DS. Moreover, the manuscript contains an overview (Table 5.3) of health care disciplines relevant to all people with DS and disciplines relevant to only few people with DS.
Some formulations are not right or precise. (e.g. contain terms like 'reasonable')	A note was added indicating that it still is to be defined what is meant by terms such as 'reasonable'.
Per specific QI (per quality dimension)	
Quality dimension: **Effectiveness, accessibility and safety**	
QIs concerning "time between onset of health problem and treatment": - Unclear how to define 'onset'. Better to take signalling the health problem as starting point. - Large differences in patient delay. Symptoms are sometimes seen as part a chronic illness and therefore untreatable.	- 'onset' is replaced by 'signalling'. - Refined: Time between first contact with a health care professional and treatment by the right professional for the concerning health problem.
QI concerning "whether guidelines are adhered to": - Or whether deviation from guidelines is well substantiated.	'or reasoned deviation' is added in the formulation of the QI.

QI concerning "DS-specific education/training": - Mention explicitly that this concerns DS-specific conditions and diagnostics. - Does this also include professional experience? - Also include ID-specialised. - This is often not feasible for all health care professionals, perhaps this QI should only apply the main/ultimate responsible professional. - Such training does not exist (yet)	- Refined: 'diagnosis, treatment or handling of DS-specific conditions, health problems and behaviour' - Refined: 'ample (to be defined) professional experience with people with DS'
Quality dimension: **Organisation, coordination and continuity**	
QIs concerning "transition from child to adult care": - Transfer of data is very important - Also other transitions are important, for instance: from living with parents to living in a assisted living facility or from assisted living facility to hospital.	A footnote ([d]) is added indicating that the formulated QIs may also be applicable to other transitions.
QIs concerning "Presence of a case manager": - Not realistic to assign a case manager for each person with DS - Key health care providers can fulfil this role - Is important	A footnote ([c]) is added indicating that the role of case manager may be fulfilled by a (key) health care professional. Furthermore, we aimed to formulate QIs ideally being part of a QI-set for health care for people with DS, regardless of any practical obstacles.
QIs concerning "Collaboration agreements": - Actual collaboration is more important than collaboration agreements. - Use terms such as 'multiple disciplines formulate one combined advice'	No change, refinements related to collaboration are applied in QIs regarding 'presence of a Downteam' (see below)
QIs concerning "ease of making referrals": - Referrals imply partitions between health care professionals, while mutual communication about care and back-referrals are important. - Add: after referral, professional to whom referral is made provides adequate feedback to the referring professional. - Not 'ease of', but 'timely' referrals.	- 'Feedback' is added in the formulation of the QIs addressing referrals. - 'ease of' is replaced by 'timely'.
QI concerning "usual source of care: Is desirable, but not always feasible.	No change, the inDicatorS-project aimed to formulate QIs ideally being part of a QI-set for health care for people with DS, regardless of any practical obstacles. In a later stage, (un)feasibility may be used as a reason to exclude the QI from further development.
QIs concerning "presence of a care plan": - Is a legal requirement - Does not always exist - Some care plans are better than others, therefore the presence of a care plan may not be a valid QI. - A care plan should be adjustable based on changed health care needs. - One care plan in which all involved professionals can add their specific information is desirable. The legal representative could be the administrator/manager of the plan.	Criteria of care plan are added: - It can be adapted in case of changed needs, - It can be accessed by all professionals involved and by the (representatives of) the person with DS. - It involves information from all relevant stakeholders.
QIs concerning "involvement of relevant stakeholders": - Should be part of the care plan	'involvement of relevant stakeholders' is added to criteria for care plan (see above)

QI concerning "presence of a Downteam within a hospital": - Is important - Is not feasible/desirable for every hospital, better to have a specialised Downteam in every region (but this may negatively affect travel distance) - Downteams should be flexible in adding / consulting specific expertise needed for specific patients - Composition of Downteams should be defined (not presence). - Use terms such as 'multiple disciplines formulate one combined advice'	- Operationalisation: 'Presence of a Downteam' is changed into: 'Sufficient (to be defined) coverage and distribution of multidisciplinary Downteams'. - QI: 'Whether a hospital has a Downteam' is changed into: 'Number and geographically distribution of Downteams that:[DS] o Have a multidisciplinary composition o Have a flexible composition based on patient's needs (possibility of removing or adding (external) disciplines to the team) o Provide people with DS / their caregivers with a written combined, harmonised, advice of all professionals involved.'
- Downteams for adults are unnecessary, every ID physician is fit/trained for the medical guidance of adults with DS, supported by other (specialised) professionals if needed).	No change, in a later stage, this may be used as a reason to exclude the QI from further development.
Quality dimension: **Person-centredness and communication**	
QI concerning "Availability of an instrument mapping the personal situation of a person with DS": - This is the most important QI - Very good idea	no change
QIs concerning "Consideration of preferences and values": - Please change: "consideration of preferences and/or values". Values are not always applicable.	'and' was replaced by 'and/or'.
QI concerning "early intervention / family support": - This QI concerns children, while other QIs concern both children and adults. - Especially relevant for paediatrician, not for other disciplines	no change, family support may be most relevant to children, but may also be relevant to adults with DS who are living with their parents.
QI concerning "whether health care professionals are trained in communication with people with DS/ID": - Is important - Should be part of each training course. - Could retaining such skills also be a QI?	no change, added: 'and keep their skills up to date'

Appendix 5-III

Longlist of potential QIs formulated for each quality operationalisation, grouped per quality dimension and quality sub-dimension

No.[a]	*Quality operationalisation to be measured by QIs*	*Potential QI(s)*	*Organisational level(s) potential QIs could apply to*	*Potential data source(s) / measurement instrument(s)*	*Type of QI? structure / process / outcome*
1	**Effectiveness**				
1.1	Effectiveness - Timely recognition of health problems: Whether health problems are sufficiently and timely recognised and treated				
1.1a	Satisfaction of people with DS / their caregivers about health / quality of life	Number of patients with DS that is satisfied about their health / quality of life after treatment.	Per health care professional Per health care organisation / practice	Patient survey Dialogue health care professional – person with DS / caregiver	Outcome

1.1b	Objective health outcomes (e.g. outcomes of blood test, physical examination)	Number of patients with DS that has improved objective health outcomes.	Per health care professional Per health care organisation / practice	EMR (automated extraction); data entry partly by health care professional, partly by others.	Outcome
1.1c	Adherence to guidelines (e.g. are recommended screenings performed and frequency of contact moments with specific professionals) or reasoned deviation	Extent to which professionals adhere to guidelines or deviate from guidelines well-reasoned.[f] Screenings suggested to be checked (based on studies 2&3 or recommended by guidelines)[b]: • Heart defects / functioning • Thyroid function (children) • Coeliac disease (children) • Hearing / ear problems • Vision / eye disorders • Dental (problems) • Dementia (older adults) Periodic appointments to be checked (based on studies 2&3 or recommended by guidelines)[b]: • Paediatrician (children) • ID physician (adults, children: if needed) • Downteam child (children) • Downteam adult (adults) • ENT-physician • Ophthalmologist / Orthoptist • Dentist • Physiotherapist • Speech therapist • Dietician • Youth care (children)	Per health care professional Per health care organisation / practice	EMR (automated extraction)	Process
1.1d	Time between signalling of health problem and treatment	I. Time between first contact with a health care professional and treatment by the right professional for the concerning health problem.	Per region Per health care organisation / practice (e.g. general practice)	Patient survey Data of health care organisation / registry	Process

		II. Number of patients with DS with one specific health problem who are satisfied about the time between first contact with a health care professional and treatment by the right professional for the concerning health problem.	Per region Per health care organisation / practice (e.g. general practice)	Patient survey	Process
1.2	Effectiveness - Expertise of providers				
1.2a	DS-specific training / education completed by health care professionals and/or their professional experience with people with DS.	Number of health care professionals having completed training / education addressing diagnosis, treatment or handling of DS-specific conditions, health problems and behaviour; and/or having ample (to be defined) experience.[f]	Per health care professional Per health care organisation / practice	HRM-data of health care organisation / registry	Structure
1.2b	Expertise experienced by people with DS / their caregivers	I. Number of patients with DS that is satisfied about the DS-specific expertise of the health care professional(s) they last visited.[f]	Per health care professional Per health care organisation / practice Per Downteam	Patient survey	Process
		II. Number of patients with DS that is satisfied about the discipline-specific expertise of the health care professional(s) they last visited.	Per health care professional Per health care organisation / practice Per Downteam	Patient survey	Process
1.3	Safety				
1.3a	Availability of safety protocols	Whether a safety protocol for providing health care to people with DS is present and is adhered to.	Per health care organisation / practice	Database/ registry of health care organisation	Structure
2	**Organisation of care**				
2.1	Organisation, coordination and continuity in general				
2.1a	(multidisciplinary) collaboration or coordination networks	I. Number of (in)formal collaboration agreements with other professionals or departments (internally).	Per Downteam (between members of team) Per health care organisation / practice (between professionals of the organisation / practice)	Database/ registry of health care organisation Inspectorate registry / audit	Structure
		II. Number of (in)formal collaboration agreements with (professionals of) other health care organisation / practice (externally).	Per Downteam Per health care organisation / practice Per region or nationally (between health care and other (non-health care) services)	Database/ registry of health care organisation Inspectorate registry / audit	Structure

2.1b	Presence of an internal case manager [c]	Number of patients with DS having a case manager who has an overview of different health care appointments, and who supports people with DS and caregivers in preparing and managing the appointments. [g]	Per health care organisation / practice	Patient survey Database/ registry of health care organisation(s)	Structure
2.1c	Presence of a (regional) case manager	I. Number of appointed case managers who are under contract of one or more of cooperating organisations, having an overview of health care and other services, helping patients finding and managing the needed health care and services. [g]	Per region or community	Database/ registry of health care organisation(s) / service(s) / regional registration Patient survey	Structure
		II. Number of people with DS having a case manager (if desired) who has an overview of different (health)care appointments and helping patients planning their appointments. [g]	Per region or community	Database/ registry of health care organisation(s) / service(s) / regional registration Patient survey	Structure
2.1d	The presence of a care plan	Number of people with DS having a care plan, that: - is based on needs of the patient, - involves information from all relevant stakeholders - can be adapted in case of changed needs, - indicates steps and time planning of treatment by different health care professionals within the organisation - indicates steps and time planning of treatment by different health care professionals and needed services (inside and outside health care). - that can be accessed by all professionals involved and by the (representatives of) the person with DS.	Per (health)care organisation	Patient survey Inspectorate registry / audit	Structure
2.1e	Whether referrals are made easily	I. Number of patients with DS that is satisfied about the convenience of referrals to other health care organisations/professionals or to non-health care services.	Per Downteam Per health care organisation / practice Per region or nationally	Patient survey	Process
		II. Number of (health care) professionals that is satisfied about the convenience of referrals to other (health care) organisations/professionals or services, and about feedback from these other organisations/professionals.	Per Downteam Per health care organisation / practice Per region or nationally	Database/ registry of health care organisations / services Professional survey	Process

2.1f	Experienced (multidisciplinary) collaboration, coordination, and continuity within the organisation	I. Number of people with DS that is satisfied about (multidisciplinary) collaboration, coordination, and continuity in one Downteam / health care organisation / practice and between all needed health care and services.	Per Downteam Per health care organisation / practice Per region or nationally	Patient survey Dialogue between health care professional / case manager and person with DS/ caregivers	Process
		II. Number of health care professionals that is satisfied about (multidisciplinary) collaboration, coordination, and continuity in one Downteam / health care organisation / practice and between all needed health care and services.	Per Downteam Per health care organisation / practice Per region or nationally	Professional survey	Process
2.2	Organisation, coordination and continuity specifically for Downteams				
2.2a	Sufficient (to be defined) coverage and distribution of multidisciplinary Downteams	Number and geographically distribution of Downteams that:[f] - Have a multidisciplinary composition - Have a flexible composition based on patient's needs (possibility of removing or adding (external) disciplines to the team) - Provide people with DS / their caregivers with a written combined, harmonised, advice of all professionals involved.	Per Downteam	EMR Patient survey, Database/ registry of health care organisation Data of health insurance company	Structure / process
2.2b	Case manager	Availability of an appointed case manager / coordinator, planning the appointments, gathering information from other health care professionals (outside Downteam / organisation), and helping patients preparing the appointments.[f]	Per Downteam	Patient survey Database/ registry of health care organisation	Structure
2.3	Organisation, coordination and continuity specifically related to transition from child to adult care[d]				
2.3a	Whether data are transferred from paediatrician to ID physician / general practitioner	The number of patients with DS aged 16-23 years having a record of data transference to ID physician or other health care professional.[g]	Per health care professional (paediatrician, ID physician or other) Per health care organisation / practice	EMR (paediatric and adult) Patient survey	Process
2.3b	Whether patients are satisfied about transition in health care	The number of patients with DS aged 16-23 years that is satisfied about the transition of health care.[g]	Per health care professional (paediatrician, ID physician or other) Per health care organisation / practice	Patient survey, Database/ registry of health care organisation	Process

2.3c	Whether patients are satisfied about transition in non-health care	The number of patients with DS aged 16-23 years that is satisfied about the transition in non-health care.[g]	Per health care professional (paediatrician, ID physician or other) Per health care organisation / practice	Patient survey, Database/ registry of health care organisation	Process
2.3d	Presence of a transition protocol	I. Whether the health care organisation providing paediatric care to children with DS has a DS-specific transition protocol, indicating information to be transferred and steps to be taken.[f]	Per health care organisation / practice	Database/ registry of health care organisation	Structure
		II. Whether the health care organisation providing adult care to adults with DS has a DS-specific transition protocol, indicating information to be transferred and steps to be taken.[f]	Per health care organisation / practice	Database/ registry of health care organisation	Structure
2.4	Accessibility – Health care nearby / within community or in primary care centres				
2.4a	Distribution of DS-specialised health care providers per region	I. The number of health care professionals using the QIs.[f]	Per region or country	National database	Structure
		II. The number of patients with DS having needed health care professionals within reasonable distance (to be defined) from their homes.	Per region or country Per Downteam	National database	Structure
		III. The number of patients with DS that is satisfied about the proximity of needed health care professionals.	Per region or country Per Downteam	Patient survey National database	Structure
2.4b	Presence of a usual source of care[e]	I. Number of people with DS having a place where they usually go when ill / in need of advice.	Per region or country	Patient survey National database	Structure
		II. Possibility of direct communication (e.g. telephone services) with that usual source of care.	Per health care professional Per Downteam Per health care organisation / practice Per region or country	Patient survey National database	Structure
3	**Person-centredness**				
3.1	General				
3.1a	Whether health care professionals are trained in person-centredness	Number of health care professionals who completed a training in providing person-centred care, or who can prove their person-centred care skills in another way.	Per Downteam Per health care organisation / practice Per region or country.	Database/ registry of health care organisations / services	Structure

3.2	Impact/burden of health care/treatment on patient's life and on his/her environment				
3.2a	Whether provider maps personal situation and adapts treatment/ advice/support accordingly	I. Number of health care professionals who made a record of having mapped the personal situation and adapted treatment accordingly.	Per Downteam Per health care organisation / practice	EMR	Process
		II. Number of patients with DS who feel that their situation is taken into account in deciding about treatment and giving (medical) advice.	Per health care professional Per Downteam Per health care organisation / practice	Patient survey	Process
		III. Number of health care professionals who made a record of having balanced the burden(s) of a treatment/intervention and the potential outcomes for the patient with DS in terms of quality of life and participation in society.	Per Downteam Per health care organisation / practice	EMR	Process
		IV. Number of patients with DS who feel that the burden(s) of a treatment/ intervention are carefully balanced against the potential outcomes for the patient with DS in terms of quality of life and participation in society.	Per health care professional Per Downteam Per health care organisation / practice	Patient survey	Process
		V. Availability of an instrument/tool enabling health care professionals to map personal situation.	Per Downteam Per health care organisation / practice	Database/ registry of health care organisation	Structure
3.2b	Health care providers make sure family support is being offered if needed.	I. Number of health care professionals who made a record of having checked the need for family support (e.g. "Early intervention" or other support). [9]	Per Downteam Per health care organisation / practice	EMR	Process
		II. Number of patients with DS who report that family support was discussed during the last consultation. [9]	Per health care professional Per Downteam Per health care organisation / practice	Patient survey	Process
3.3	Involvement of all relevant stakeholders				
3.3a	Whether providers involve patients/ caregivers/other providers in decisions	I. Number of health care professionals who made a record of having involved all relevant people for the decision at stake.	Per Downteam Per health care organisation / practice	EMR	Process

		II. Number of patients with DS who feel that all relevant people for the decision at stake are involved.	Per health care professional Per Downteam Per health care organisation / practice	Patient survey	Process
3.4	Consideration of preferences and values of the person with DS and his/her family				
3.4a	Whether person with DS / caregivers feel their values/ preferences/ worries are taken into account.	Number of patients with DS that is satisfied about the extent to which a (health care) professional takes the patient's values/preferences/worries into account.	Per health care professional Per Downteam Per health care organisation / practice	Patient survey Dialogue health care professional – person with DS / caregiver	Process
3.5	Communication: Whether provider adapts communication to (dis)ability of patient and build a trust relationship				
3.5a	Satisfaction of patient / caregiver regarding communication and trust relationship	I. Number of patients with DS that is satisfied about communication during last consultation.	Per health care professional Per Downteam Per health care organisation / practice	Patient survey	Process
		II. Number of patients with DS (or their caregivers) that understands information provided during consultation.	Per health care professional Per Downteam Per health care organisation / practice	Patient survey	Outcome
		III. Number of patients with DS (or their caregivers) experiencing a trust relationship with the health care professional most visited.	Per health care professional	Patient survey	Process
3.5b	Whether health care professionals are trained in communication with people with DS/ID	Number of health care professionals who completed a training in communication with people with DS/ ID, or who obtained their communication skills in another way, and who keep their skills up-to-date.	Per Downteam Per health care organisation / practice Per region or nationally.	Database/ registry of health care organisation	Structure

Applicable to both children and adults if not specified otherwise
Abbreviations: DS=Down syndrome; EMR=electronic medical record; ENT=Ear Nose Throat; ID=Intellectual disability/ies; HRM=human resource management; No.=number; QI=Quality Indicator.
[a] Number indicating quality dimension (for example: 1), quality sub-dimension (for example: 1.1), and quality facet (for example 1.1a).
[b] Study 1: literature review on existing QIs for health care for people with DS (van den Driessen Mareeuw et al., 2017); Study 2: Qualitative exploration (interviews and focus groups) among people with DS, their parents and support staff (unpublished work by the authors); Study 3: Delphi-study among health care professionals and patient organisations' representatives (unpublished work by the authors). Dutch guideline on health care for children with DS (Borstlap et al., 2011) and the preliminary stage of the Dutch adult guideline (which is currently being developed).
[c] The role of case manager may be fulfilled by one of the (key) health care providers of the person with DS.
[d] Formulated QIs may also be applicable to other transitions, such as: from living with parents, to living in assisted living facilities, or from assisted living facility to hospital.
[e] Usual source of care: after Coker et al, 2012 ("National Survey of Children's Health" (USA))
[f] Relevant to people with DS only
[g] Relevant to people with DS and other people with complex disease.


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Good health care for a good life? The case of Down syndrome

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ABSTRACT

People with Down syndrome have complex health care needs which are not always fully met. Health care improvements are required in order to better meet these needs. Quality indicators are an important tool for improving health care. However, quality indicators for health care for people with Down syndrome are scarce. Existing quality indicators focus on medical (physical) needs or the clinical setting, even though it is acknowledged that quality measures should reflect the *total* of quality aspects relevant to the population at stake, which may encompass aspects *beyond* the medical domain. These aspects beyond the medical domain are the focus of the current paper, which aims to provide insight into the way people with Down syndrome live their lives, how health care may fit in, and how this may impact the development of quality indicators.

The paper is based on data originating from interviews with people with Down syndrome and their parents as well as focus groups with support staff members working in assisted living facilities for people with intellectual disability.

The data revealed a lot of variation in how people with Down syndrome live their lives. Nevertheless, we were able to identify eleven topics, which we grouped into three overarching themes: 1. Being different yet living a normal life; 2. Down syndrome-(un)friendly society and services; and 3. Family perspective.

The variation in our data stresses the importance of health care that takes a person's life into account beyond the medical domain, as exemplified by the identified topics. Our findings also show that a good life is not merely depending on good health care supported by well-defined quality indicators, but also on (support in) all life domains.

INTRODUCTION

Down syndrome (DS) is a common cause of intellectual disability (ID) (De Graaf et al., 2017; Phelps et al., 2012), and is related to a specific combination of health problems, behavioural profiles, and cognitive challenges (Capone et al., 2018; Capone et al., 2020; Coppus, 2017; Grieco et al., 2015; Weijerman & De Winter, 2010). Because of this, people with DS have complex health care needs, which are not always properly met by the health care they receive (Capone et al., 2018; Goodman & Brixner, 2013; Peters et al., 2020; Phelps et al., 2012). A complete picture of what matters to patients is required to properly answer their needs (Czypionka et al., 2020; Kelley & Hurst, 2006) and may include quality aspects *beyond* the medical domain (Czypionka et al., 2020). This is supported by our previous work that showed that parents of people with DS consider health care as only one aspect of the total of desired services and facilities (Van den Driessen Mareeuw et al., 2020b).

Quality indicators (QIs) may largely contribute to obtaining such a complete picture of what matters to patients and to improving health care (Donabedian, 2005; Porter, 2010). Current developments in health care quality measurement underline the importance of measures that matter to patients (Kelley & Hurst, 2006; Porter, 2010) and that reflect the *total* of quality aspects relevant to the population and context at stake (Terwee et al., 2018). It is acknowledged that the social environment (family members, other caregivers, house mates) and the contexts in which people with ID (including people with DS) live, may all interfere with outcomes of health care (Goodman & Brixner, 2013; Kyrkou, 2018; Mastebroek et al., 2016; Simões & Santos, 2016). This is also in line with the currently increased attention to person-centred care and related shared or collaborative decision-making (Peisah et al., 2013; Poitras et al., 2018).

Despite the acknowledged importance of a broader perspective (Czypionka et al., 2020; Kelley & Hurst, 2006), most quality improvements related to health care for people with ID are focused on medical (physical) needs or the medical/clinical setting (Van den Driessen Mareeuw et al., 2017; Jespersen et al., 2018). In addition, existing QIs either cover medical care for people with ID in general, without specifically addressing certain conditions or treatment courses, or cover the support and care available in supported living facilities (Van den Driessen

Mareeuw et al., 2017). The general nature of these existing QIs on the one hand and the lack of QIs covering the complete picture of what matters to patients on the other, urges for the development of DS-specific QIs, as these are currently almost non-existent (Van den Driessen Mareeuw et al., 2017; Santoro et al., 2021).

The Dutch inDicatorS-project was set up to develop QIs for health care for people with DS that are sensitive to their specific needs. This paper aims to provide insight into the way people with DS live their lives, how health care may fit in, and what this means for the development of QIs. The following research question is addressed:

What is important in the lives of people with DS, and how could this impact the development of QIs for health care for people with DS?

METHODS

This paper is based on data from semi-structured interviews with people with DS, semi-structured interviews with parents of people with DS and focus groups with support staff working in assisted living facilities for people with ID (including people with DS), which were conducted as part of a qualitative explorative study on health care quality from the perspective of people with DS and their caregivers (Van den Driessen Mareeuw et al., 2020b). The study meets ethical guidelines and legal requirements.

Participants

As described before (Van den Driessen Mareeuw et al., 2020b), purposive sampling was applied to ensure a large diversity of participants and obtain insights from different perspectives. Inclusion criteria for participants with DS were being able to participate in an interview, and therefore being at least twelve years of age, and to have mild to moderate ID. Because of their significant role in the lives of people with ID (Mastebroek et al., 2016), parents and support staff of people with DS were involved to obtain complementary information about people with DS, but also to obtain information about people with DS who are not able to participate in an interview (younger than twelve, or with more severe ID).

Participants were recruited through the Dutch DS association, service providers for people with ID, and the network of the authors. All participants received participant information and informed consent forms; participants with DS received easy-to-read versions. Participants, and their legal representatives if required, gave informed consent.

Data collection

Semi-structured interviews with people with DS and with parents

The interview protocol for the interviews with people with DS and with parents consisted of an introductory part, including information about the study and about participation in an interview, and a list of topics to be discussed, although the detail and order in which the topics were addressed differed per participant group. The topics included experiences with health care for people with DS and topics derived from the eight domains of quality of life by Schalock et al. (2005): Emotional well-being, interpersonal relations, self-determination, social inclusion, material well-being, personal development, rights, and physical well-being. Furthermore, participants were allowed to add topics they considered relevant. The interviews were conducted by one of the authors.

Participating people with DS and parents could choose the time and venue of the interview and were allowed to invite someone else for moral and/or verbal support. The abilities of the participants with DS were met by using visual materials, such as pictures of care settings, daily activities, and pictograms reflecting emotions and other abstract concepts. Furthermore, the interviewer's talking pace and phrasing was adapted to the abilities of the participant with DS, and extra time was dedicated to putting the participant at ease. Such (adapted) interviews are often used in research involving people with ID and generally result in sufficient data (Frankena et al., 2015).

Focus groups with support staff

The protocol for the focus groups with support staff was similar to the interview protocol in terms of topics discussed, but differed in terms of detail and order in which topics were discussed and attention paid to group work (e.g. listening to each other, not talking at once). The focus groups were convened by the author who also conducted the interviews.

Five focus groups with five to twelve support staff members took place in meeting rooms of the service provider where participating support staff were employed. One support staff member was interviewed individually, because he was not able to join the focus groups.

Data analysis

Data saturation occurred in both interviews and focus groups, meaning that additional interviews or focus groups did not yield new relevant information (Tong et al., 2007).

Pseudonymised transcripts were made of the audio recordings of the interviews and focus groups. Data analysis was done using the software package Atlas.ti 8 for Windows, and consisted of three steps, based on the framework analysis method (Gale et al., 2013):

1. *Deductive and inductive coding (Gale et al., 2013).* Text fragments of first few transcripts were labelled with codes indicating relevant information. Deductive coding included predefined codes based on quality of life domains (Schalock et al., 2005), dimensions of quality of care (WHO, 2006), and principles of patient centred care (Singer et al., 2011), allowing structuring of data. Inductive coding involved open codes, formulated based on the content of text fragments, ensuring that no themes were missed.
2. *Constructing and applying an analytical framework*. One third of the transcripts was double coded by two authors (one ninth by authors 1 & 2, authors 1 & 3, and authors 1 & 4). The analytical framework was evaluated by comparing and discussing the attributed codes by the different authors, after which it was adapted (merging, splitting and sorting codes per theme), leading to a final framework which was applied to the other transcripts.
3. *Charting data*. For each code within the themes, text fragments were summarised and put in a framework matrix, allowing data interpretation. The matrix differentiated between perspectives from people with DS, parents, and/or support staff.

This resulted in themes including codes related to medical/health care aspects, but also in themes and codes related to broader issues, providing insight into the lives of people living with DS and their families. The data retrieved on these broader issues are the basis of the current paper. The data retrieved on the

medical/health care aspects are published elsewhere, as well as more information on data collection and analysis (Van den Driessen Mareeuw et al., 2020b).

RESULTS

Eighteen people with DS (ten female, eight male) participated in the study, ranging from twelve to 54 years old, living with their family (4) or in assisted living facilities (14), and had mild to moderate ID (Van den Driessen Mareeuw et al., 2020b). Fifteen parents (or parent couples) of people with DS participated in the study. Their children ranged from two to 43 years of age (7 female, 8 male) and lived with their family (11) or in an assisted living facility (4), and had severe to borderline ID. A total of 35 support staff members participated in the study. Their clients with DS ranged from 24 to 63 years of age and had severe to borderline ID. Further details about participants can be found elsewhere (Van den Driessen Mareeuw et al., 2020b).

The data providing information on the lives of people with DS resulted in three themes containing a total of eleven topics:

Theme A - "Being different yet living a normal life", topics:

1. Activities
2. Work/School
3. Social relationships
4. Housing
5. Barriers and levers for a normal life

Theme B - "DS-(un)friendly society and services", topics:

6. Societal inclusion and image of people with DS in media
7. Autonomy
8. Services and support
9. Balance between autonomy and healthy choice

Theme C - "Family perspective", topics:

10. Arranging help, support, and services
11. Impact of having a child with DS

The topics are described in three paragraphs corresponding to the overarching themes. For each topic, examples are provided by means of quotes from the participants.

A - Being different yet living a normal life

Participants with DS indicated that they wanted to be just like others, including having an apartment of their own, having a job, having a partner and friends, and being independent. A mother (55yrs) illustrated the desire of her daughter (22yrs) to be like anyone else as follows: *"She really wants to be independent. […] She sees her sisters leaving the parental home, going on holiday on their own, going out with friends. Well, she also wants that, you know."*

1. Activities

The participants with DS showed a varied picture of how they lived their lives. Activities mentioned by participants with DS included: school, internships, work, or activities in an activity centre for people with ID, and a large variety of hobbies, such as sports, acting, painting, musical activities, etcetera. For example, a woman with DS (54yrs) described her activities for the coming Saturday as follows*: "Tomorrow I'll go for a swim, and when I'm home I'll drink coffee, and after coffee I take a shower, and then I'm going to a birthday party, of a friend."* And for weekdays: *"I'm at work during the day".* Parents and especially support staff described the lives of people with DS as quite busy. This support staff member (woman, 55yrs) described: *"If I look at my two downers [clients with DS], well, they are really having a busy life, full of all kinds of nice activities."* Support staff and parents indicated that some people with DS even become overcharged with activities or are confronted with too high expectations (e.g. by parents). This mother (55yrs) of a daughter with DS (23yrs) illustrates these expectations: *"They've got this syndrome you know, but they all have to become like us, so I think: how is that possible?".*

2. Work/school

During the weekdays, activities of adult participants with DS ranged from having one or more jobs, often in sheltered workshops, to activities in activity centres for people with ID. Generally, the participants with DS valued and liked their jobs or activities and their colleagues. A woman with DS (39yrs) mentioned that by having a job "*We are showing that we're also there and […] that we can also do this".* A woman with DS (23yrs) points out: "*sometimes we are going for a bite with my colleagues*".

School-aged children with DS either went to a specialised school for children with ID or to a regular school where they often received extra support by specialised staff. Parents' stories were varying about specialised education. Some mentioned

that the quality of specialised education was low, while others reported that the switch from regular to specialised education was relieving, because the specialised school better matched the abilities of their child.

3. Social relationships

Participants with DS regarded (sometimes deceased) parents, siblings, other family members, and support staff as the most important people in their lives. For example, a woman with DS (41) said: "*I sometimes go to grandma by myself.* Her mother, who joined the interview added: *"to the graveyard"*. Another woman with DS (54) indicated the people most important to her*: "sometimes the support staff here [in her living facility], but my brothers the most*"(…) *"I'm happy that I still have my brothers, and my sisters in law, and my cousins".* A wide range of other people including friends, boyfriends/girlfriends, colleagues and house mates, but also frequently visited health care providers were mentioned as important. Parents and support staff confirmed this. However, parents also noted that people described as friends by people with DS, are often friends of siblings or parents.

Opinions about the desire to have a boyfriend or girlfriend varied largely among the participants with DS. Some had one or were longing to have one, whereas others did not have one or preferred friends over a boyfriend/girlfriend. A woman with DS (32yrs): "*In the past, I did have a boyfriend, but now I want to stay single*". In looking for a partner, this woman with DS (28yrs) also expressed a desire to be just like others: *"So they [dating service for people with ID] are trying to find one [a boyfriend] for me… And I said: … if only he is normal, only has a slight handicap, not a severe one.".*

4. Housing

Experiences with housing also varied among the participants. Some people with DS were quite happy with where they lived. A woman with DS (41yrs) described her assisted living facility as follows: "*like happiness".* However, others felt lonely or otherwise unhappy with where they lived. For example, a woman with DS (28yrs) who lived in an assisted living facility, revealed that she was afraid of becoming lonely there and preferred her parental home: *"I'm afraid of loneliness (…) but not here [at her parents' home]".* Participants with DS who were living with their parents either preferred to stay in this situation or were excited to be 'leaving home' in the near future.

5. Barriers and facilitators for a normal life

Even though being just like everybody else is important for people with DS, participants acknowledged, sometimes with frustration, that having a normal life was sometimes hindered by their cognitive or physical conditions related to DS and that they needed support. A man with DS (32yrs) working in a hotel run by people with ID told about his work: "*I do work independently. But I do need guidance.*" Both parents and support staff explained that communication problems (e.g. speaking/hearing problems, slow information processing) impede social interaction. A mother (57yrs) of a son (25yrs) with DS illustrated: "*That is because he is slow, also compared to other persons with Down [syndrome], he is slow. Other people with intellectual disabilities often react much quicker and then they are finishing his sentences and he doesn't want that*". Additionally, support staff (parents to a lesser extent) brought to light that around the age of forty, people with DS are becoming less active, possibly as a result of early ageing. A support staff member (woman, 26yrs) noted: *"And I also see diminished initiative"*.

Parents (sometimes siblings) and support staff offer the needed support for achieving a life as normal as possible. They provide emotional support, but they also accompany people with DS in (health) care appointments, arrange transportation to hobbies/other activities, and manage social contacts. Other mentioned examples of support offered by parents include: the formation of a group for children preparing them for school, sometimes even fulfilling the role of a schoolteacher, supporting development or arranging needed support, being involved in setting up specialised medical services, and creating sports groups for people with ID.

B - DS-(un)friendly society and services

Participation in society and autonomy were also considered important elements in the lives of people with DS. Although people with DS generally feel they are part of society and that their autonomy is respected, they may encounter difficulties and need extra support in these areas, as society and services may not always be DS-friendly.

6. Societal inclusion and image of people with DS in media

Generally, people with DS gave the impression that they felt part of society. However, people with DS, parents and support staff also reported that people with DS felt lonely, were being bullied or not accepted because of their DS. For

example, a woman with DS (38yrs) said: *"In the past, I was bullied at school. […] They used this other word for syndrome, they were calling me 'mongol', but I'm not a mongol, I'm just myself and I have Down syndrome. […] But it was not easy."* A mother (49yrs) of a son (13yrs) pointed out: *"He [son] and this boy really had a click. This boy did not have Down syndrome, but he was at that school for a reason, had a low IQ. They wanted to make an appointment to play together, but it was never possible, I didn't know why, and finally it became clear that his mother could not accept that her son had a friend with Down syndrome and she prevented appointments."* A support staff member (woman, 59yrs) revealed that other people with ID were sometimes not accepting people with DS because of their specific appearance: *"Down syndrome is quite visible, I think that some of the others [without DS] who live here [in assisted living facility] do not want to be seen with someone with Down syndrome"*. It was also noted that people in the street do not always know how to approach people with DS. This mother (57yrs) of a man with DS (25yrs) explained: *"People do not know how to handle [name son] and I can't blame them for that. So other people observe how we act, […] they do as we do. For example, the hairdresser is also trying to do what we do, and that's so really nice to see. […] But we have to set an example, because people do not know what to do".*

Furthermore, parents as well as support staff indicated that often an unrealistic or stereotypic image of people with DS is presented in the media, only showing people with DS who are quite independent and participate in society quite well and like to be in the centre of attention. A support staff member (man, 44yrs) explained: *"This is what you see on TV, they all want to be on stage, they all want to grab the microphone, and being in the centre of attention"*. Parents argued that this would negatively impact the societal feeling of urgency in providing support for people with DS. For instance, a mother (63yrs) of a daughter (28 yrs) with DS argued*: "In response to [names of presenters of Dutch TV-shows involving people with DS], a medical doctor wrote in the newspaper that it was just as if it's a pity if you don't have a child with DS. Well, of course, it's not like that, you know. […] Our daughter always needs support and guidance".*

7. Autonomy

The interviews with people with DS showed that they generally have freedom of choice, or are at least involved in decisions regarding housing, daily activities, work, etcetera. A woman with DS (54yrs) illustrated that she may decide herself where

she wants to have dinner: *"If I want to eat upstairs. Sometimes I don't want to eat with the others in the common room, when they are all arguing and all. I cannot stand that."* In some cases, a feeling of autonomy was created by parents or support staff, for example by letting the person with DS do the talking with health care professionals during consultation, only intervening (non-verbally) when necessary, and supporting people with DS in making their own decisions. Sometimes, in the best interest of the person with DS, they pretend the person with DS makes his/her own choices. A support staff member (woman, 26yrs) explained: *"I try to make it look like as if it is their own choice, while it is also the right choice, or that they can choose between two right options".*

8. Services and support

Independence, autonomy and inclusion in society was much stimulated by all kinds of different services and support systems, by developmental support (in young children with DS) and "*activities that stimulate them, so they have to do more than only assembling screws, so like gardening, shopping and planning that, musical therapy*." (Support staff member (woman, 26yrs)). However, there were also cases in which people with DS had to live according to the system with little room for making their own choices. For example, the mother of a woman (28 yrs) with DS unveils: *"She is always dependent and she always has to do as she is told. She has to go to bed when she is told to do so […], she has to eat what is served. […] If the group is going to the funfair, she has to join them, whether she wants it or not, because she cannot stay at home alone"*.

9. Balance between autonomy and healthy choice

Health was promoted in all kinds of ways in order to improve life and participation. All participants with DS were well aware of the positive impact of medication, (medical) aids or support, such as physiotherapy, a walker, and arch support for better walking. Parents added that speech therapy, contributing to better communication skills, was especially important at a younger age. Lifestyle, especially being overweight and on a diet, was often an issue among people with DS. However, despite various medical problems, participants with DS considered themselves to be healthy. It also became clear that it was not always easy to find the right balance between autonomy and personal values and ideas on the one hand, and making the healthy choice on the other. Ideas of participants with DS about health care ranged from considering it as part of their regular schedule, to

finding it tiring or not nice. Some people with DS who had a family member who had died in hospital, had developed the idea that when you go to a hospital you will die. People with DS, parents and support staff showed that (health) services and supports sometimes succeeded to find the right balance, sometimes not. For example, a woman with DS (54yrs) said: *"I may eat a bit, but not too much"*. Support staff (woman, 26yrs) of this woman explained: *"She has lost 20 kilos already. […] she's got a list of what she's allowed to eat, […] If she does well, […] she gets a reward, like doing something nice together."* A man with DS (32) said: *"Yes, I've got arch support [foot correction]"*; His mother (65yrs) who joined the interview however added: *"Yes, he had, but he threw them [insoles] away"*. Another mother (55yrs) explained about her daughter (23yrs): *"they'll say that she has to have glasses, but she just doesn't want them and she functions well, so let it go"*.

C - Family perspective

A child (or sibling) with DS may bring joy as well as worries to a family. Primarily parents noted the efforts needed to arrange all needed supports and services for their child with DS. Some parents manage quite well, whereas others experience the efforts as distressing.

10. Arranging help, support, and services

Parents play a crucial role in managing and arranging all help, support and services needed for a good life of their children with DS. All interviewed parents mentioned problems related to this. Parents reported difficulties in identifying the needed and available services for their child in their region. They questioned what day care (for young children), developmental support, (support at) school, work, housing or other activities their child needed. A mother (41yrs) of a boy (2yrs) with DS illustrated: *"What do you choose, […] which development method?"* A father (63yrs) of a woman (32yrs) with DS illustrated: *"She needed an internship when she had finished school, or work that she could do. And then you go to the municipality and they say: we don't know, maybe you can get some support here and there. We had to find out ourselves."*

Once parents had found the right (combination of) services and supports, they encountered problems in actually arranging them. They for example faced problems concerning availability of services or housing. A father (63yrs) of a man (32yrs) with DS exemplified: *"All assisted living facilities and initiatives are full, so you're dependent on available places."* Other problems were related to the efforts

needed to (financially) secure all services their child with DS was entitled to. A mentioned complicating factor was that rules and regulations were changing regularly and that different municipalities applied different rules. A mother (37yrs) of a girl (7) with DS illustrated*: "How you have to apply for all the services, that's a hell of a job. […] you have to invent the wheel yourself, […] it differs largely per municipality. […] And then you think you have arranged it all for one year, but then you have to do it again for the next. […] and then we got this discussion about whether the municipality was financially responsible or whether it was covered by some other regulation."* Additionally, parents indicated that it was quite complex to align the needed support and services, and for example arrange transportation from and to the different services. Some parents indicated that they got assistance with aligning all support and services from a local case manager appointed by one of the organisations that provided support to their child. A father (54yrs) of a boy (14yrs) illustrated: *"Well we put a lot of energy in that, and someone from the care organisation who was responsible for the guidance of the childminders, took the first step in aligning all these separate activities: speech therapy, physiotherapy, floating support, educational support at school, to bring them all together, and to make sure that we all had one goal for him [son with DS] at school and after school."*

The interviewed parents indicated that not all parents are capable of tackling the above problems and noted that some become overburdened with it. A mother (41yrs) of a boy (2yrs) explained: *"We can manage, but parents who are not that assertive, not that capable of investigating all options…"* Furthermore, it was mentioned that all activities require more time with a child with DS and that even when a child does not live with his/her parents anymore, many tasks are still to be fulfilled by the parents, such as cleaning the apartment of their child with DS and regulating the weight of their children. Parents (both 64yrs) of a daughter with DS (28yrs) explained: "*and we still have to do the rest. […] actually, I'm busier now [since daughter moved out], but in a different way, because if she's tired, she cannot do anything. She may say that she can do the washing, but that's not totally true of course."*

11. Impact of having a child with DS

The impact of having a child with DS was also an issue often mentioned by parents. On the one hand, parents indicated that their child with DS made them and other family members live 'in the moment'. For example, a mother (57yrs)

of a man (25yrs) with DS explained: *"My eldest sometimes said: If I feel stressed or not that well, then it helps me to watch [name son with DS]".* On the other hand, parents revealed that they had had difficulties accepting the diagnosis 'Down syndrome' and that they sometimes found it confronting to see children of the same age without DS or to face information about DS. They also added that they had learned to live with it. A mother (49yrs) of a boy (13yrs) with DS illustrated: *"fortunately, you get used to it, […] but my niece who is two years younger than [name son with DS], that's quite confronting. Then I think, shit, she can do this, she can do that, all independently."* Additionally, parents noted that siblings of people with DS are sometimes forgotten because all attention goes to the child with DS.

Some parents were quite worried about the future of their children with DS, while others were confident that they had made, or would make, the right arrangements for the future. Worries often had to do with their children moving out, or with themselves not being there anymore. For example, a mother (55yrs), of a woman (23yrs) with DS said: *"Yes I'm worried, whether she will get the attention she needs, when she is going to live there [in an assisted living facility]"* Another mother (63yrs) illustrated: *"All parents are bothered with this: what if we cannot do it anymore?"* Parents made several arrangements for their children, ranging from building social networks to establishing legal arrangements. For example, a father (63yrs) of a man (32yrs) with DS said: *"We have this social network around him, partly paid by this regulation, and then there is family living nearby […] so if we fall out, he will be known and recognised in our village".* A mother (63yrs) of a woman with DS (28yrs) added: *"two legal representatives […] and we're currently drafting a will".*

DISCUSSION

This paper provides insight into how people with DS in the Netherlands live their lives, how their lives are supported, and what this may mean for their parents and other family members. This insight draws the broader context within which the development of quality instruments for health care for people with DS, such as QIs, should take place (Kelley & Hurst, 2006).

This paper shows aspects of the life of people with DS that may directly or indirectly interfere with health care. An example of a direct connection to health

care can be found in the given examples regarding medical aids, such as arch support or glasses. This involves finding the right balance between autonomy and personal values and ideas on the one hand and making the healthy choice on the other. From the medical point of view, medical aids may be a good idea ('healthy choice'). However, if a person with DS does not accept the aids or is not experiencing a functional problem ('autonomy, personal values and ideas'), this may not be the best option. Therefore, before describing aids, it would be helpful to investigate whether the person with DS (and his/her carers) accepts such aids, which guidance may be needed, and whether alternatives are available. Another example is the aspect of 'wanting to be just like others' – which was considered to be quite important by the study participants, but also by other people with ID (Sandjojo et al., 2019). Its importance is also reflected as a *right* to be like others in the Convention on the Rights of Persons with Disabilities (CRPD) (UN, 2006). One participant with DS in the current study wanted to deploy the same activities as her siblings without DS and another explained that by having a job she showed that she was just like others. If accepting medical aids would enhance the feeling of being different, and not being as others, this could be a reason for refusing such aids. Health care professionals should take such issues and desires into consideration in order to contribute to the quality of life of their patients with DS. By doing this, they would also respect the CRPD (UN, 2006), which advocates support needed to establish equity.

Achieving the right balance between 'autonomy/values/ideas' and the 'healthy choice' demands a person-centred approach (Langberg et al., 2019; Morgan & Yoder, 2012; Poitras et al., 2018). Person-(or patient) centredness is multi-faceted but is generally built upon three overarching elements including the person's situation, the professional-patient/person relationship and coordinated care (Langberg et al., 2019; Singer et al., 2011), which are also reflected in the findings of this study. In many literature, person-centredness also involves shared decision-making practices, in which health care professionals collaborate and share responsibilities with their patients and the people around them in order to find the option that best fits the preferences, values and context of the patient (Langberg et al., 2019; Peisah et al., 2013). Our data show that people with DS are able, to a certain extent, to act and decide autonomously, and that their parents, other family members and support staff support them. Such 'collaborative decision-making' practices (Peisah et al., 2013), as part of a person-centred approach, will

also include and respect the situation of parents, which may be, as our findings show, quite challenging. The same holds for most parents and families having a child with ID (Staunton et al., 2020). Taking into account such contextual factors may positively impact health outcomes (Poitras et al., 2018). Moreover, by sincerely seeing and listening to people with DS, their autonomy is respected (Peisah et al., 2013), which is important because it contributes to a feeling of being seen and a feeling of being 'just like others' (Sandjojo et al., 2019).

Having said this, our findings also show that there is a lot of variation between people with DS. Although specific health problems, behavioural profiles and cognitive challenges are more common among people with DS (Capone et al., 2018; Capone et al., 2020; Coppus, 2017; Grieco et al., 2015; Weijerman & de Winter, 2010), each person with DS is unique. By striving for as much variation in the participant characteristics as possible (Van den Driessen Mareeuw et al., 2020b), we attempted to capture as many different meanings, impacts, and perspectives as possible. Despite this, we could not avoid underrepresentation of people with DS with severe ID in our study population, which may have introduced some bias. Perhaps, applying additional methods especially suited for people with limited verbal skills would have diminished this (Frankena et al., 2015). We did, however, include several parents and support staff from people with DS with severe ID. Nevertheless, the richness of the data and the broad range of insights we were able to unveil, underlines again the importance of looking into a person's life, beyond the medical domain, in order to provide effective health care and establish 'QIs that matter to patients'. At the same time, the data also show that a good life is not merely depending on good health care, but that it involves all life domains (Schalock et al., 2005). This not only means that health care professionals should respect all these domains and look for collaborations with other domains, but also that professionals, and informal carers, from all sectors should collaborate and seek for joint initiatives to support people with DS in living their lives. In fact this calls for a more supportive society, in which all people, including people with DS, can participate in their own specific way. As part of this, and in accordance with the CRPD (UN, 2006), families should be supported in the care for their family member with DS, for example by investing in personal coordinators. Especially since our findings acknowledge the key role of the family in enabling people with DS to participate in society. Extra family support could alleviate the struggles families experience with respect to arranging all services and supports needed

for a 'normal life' and participation. Similar issues are seen in families with a child with ID, as well as the need of good family support, which is not always sufficient (Staunton et al., 2020).

The findings of this study put QIs for health care for people with DS in a wider perspective. QIs are not the panacea for a supportive society, they can however contribute to it if they are developed in harmony with this wider perspective and as part of a larger whole. This implies that the QIs should reflect this wider perspective and should not only cover medical measures (e.g. whether a timely cardiac ultrasound took place), but should also include aspects related to coordination, collaboration and person-centredness. QIs may for instance use electronic health insurance claim data to measure coordination and collaboration (Uddin et al., 2015). Such QIs may stimulate health care professionals to synchronise provided care with the person's life and his/her social and institutional context. In that sense, QIs contribute to improving health care (Donabedian, 2005; Porter, 2010) and thereby to better lives of people with DS. This 'outward-looking' approach of health care (professionals), which may be stimulated by QIs, might also have a positive effect on health care for people with ID without DS, especially since previous research showed that QIs on health care for people with ID are scarce or cover other services than medical ones (Van den Driessen Mareeuw et al., 2017). Consistent with the current findings, the medical domain concerning people with ID does not seem to be connected with other services. A more 'outward-looking' and holistic approach of professionals in health care for people with DS, as stimulated by QIs for health care for people with DS, might set an example for health care for people with ID.

Conclusion

In an era in which health care and QIs ought to matter to people, a broader perspective, beyond the medical domain, should be applied. This study shows what this may encompass regarding people with DS as it provides elaborated insight into the lives of people with DS. QIs for health care for people with DS should reflect this broadness in order to contribute to their lives and should be introduced as part of a larger system, fostering, among other things, person-centredness and intersectoral collaboration. The findings may also apply to quality of health care for other people with ID.


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General discussion

Quality of health care is widely studied and improvements in health care are continuously implemented, in an attempt to improve health outcomes and to diminish costs (Amalberti et al., 2018; Busse et al., 2019). Quality indicators (QIs) are considered an important instrument in health care improvements, as the insight they provide into health care quality may inform health care reforms, health care provision and patient choices (Boulkedid et al., 2011; Campbell et al., 2002; Donabedian, 2005; Rademakers et al., 2011). However, initiatives aiming to improve health care for people with Down syndrome (DS) are scarce and QIs scarcely existent. This is remarkable since DS is associated with a large variety of typical health problems and health care is of vital importance to most people with DS (Kinnear et al., 2018; Kyrkou, 2018). Therefore, the objective of this thesis was to draft QIs that measure quality of health care for people with DS and that are sensitive to their specific needs and contexts.

The thesis includes five studies. In the first (chapter 2), it was investigated whether QIs for health care for people with DS did already exist, using a scoping review. This study concluded that such QIs are scarce; only one QI measuring the prevalence of thyroid disease among people with DS in the UK was found. This meant that QIs for health care for people with DS had to be developed from scratch. Because of this scarcity, the scope of the study was broadened to searching for QIs for health care for people with ID in general. Although the thirteen identified QIs or QI-sets were not directly applicable to health care for people with DS in the Netherlands, they informed the further development of the QIs for health care for people with DS. The development started with a qualitative explorative study, including interviews and focus groups with people with DS, parents of people with DS, and support staff working in assisted living facilities for people with intellectual disability (ID) (and DS) (chapter 3), which identified important elements of health care quality. These elements, which were mostly related to multimorbidity, well-coordinated and findable services, person-centredness, and provider-patient communication, were presented to health care professionals working with people with DS and patient organisations during a Delphi-study (chapter 4). Participants in the Delphi-study agreed upon two purposes for the QIs and upon quality issues to be measured by the QIs. It became clear that the QIs should cover a large diversity of clinical and other quality domains and should involve many health care disciplines. The study also yielded insight into preconditions and considerations for further development and use of the QIs. It was for example

unveiled that study participants feared that public quality information on the level of individual health care professionals would lead to unfair judgement of the professionals and long waiting lists, which would harm health care quality. In the fourth study, the findings of the three previous studies were brought together resulting in three main quality dimensions (i.e. effectiveness, organisation of care, and person-centredness) and a total of ten sub-dimensions (chapter 5). For each sub-dimension, potential QIs were drafted, resulting in a longlist of 46 QIs. The fifth study (chapter 6) draws upon the data collected during the qualitative exploration (chapter 3), with a focus on how people with DS live their lives. This provides contextual knowledge putting the drafted QIs into perspective.

In the following, the main findings of this thesis are presented, and the answers to the research questions are discussed. Subsequently, strengths and limitations of this thesis are discussed, after which further reflection on the findings are described. This chapter ends with future steps and recommendations.

Main findings and answers to research questions

Quality of health care for people with DS and how to measure it (Research questions 1 & 2)

A longlist of 46 potential QIs was drafted, based on three main quality dimensions and their sub-dimensions, which are shown in box 7.1.

Box 7.1 Quality (sub-)dimensions for which QIs were drafted

1. Effectiveness
   a. Timely recognised and treated health problems
   b. Expertise of professionals
   c. Safety

2. Organisation of care
   a. Organisation, coordination, and continuity:
      - In general,
      - for Downteams, and
      - related to transition from paediatric to adult health care
   b. Accessibility

3. Person-centredness
   a. General
   b. Impact/burden of treatment
   c. Involvement of relevant stakeholders
   d. Considering preferences & values
   e. Communication and trust

The identified quality (sub-)dimensions resemble quality dimensions described in the literature (IOM, 2001; WHO, 2006; WHO, 2018) and the dimensions covered by existing QIs for people with ID (chapter 2). The quality dimensions reflect the perspectives of health care professionals as well as people with DS and their caregivers. Most quality issues were mentioned by both. However, health care professionals tended to focus more on issues related to effectiveness, such as performing the right screenings, and providing the right care in the right manner (chapter 4). In contrast, people with DS, parents and support staff focused on quality issues related to person-centredness and organisation of care, such as creating a respectful trust relationship between the person with DS and the health care professional, applying a holistic approach and coordinating care services (including services outside the medical domain) (chapter 3). This perspective of people with DS and their caregivers echoes that most experienced problems were related to person-centredness, communication and organisation, while effectiveness of medical care was generally taken for granted by (caregivers of) people with DS. This may be a logical result of the large number of health care professionals and settings people with DS and their caregivers encounter, which may make them more perceptive for problems in general and specifically for problems concerning organisation. Health care professionals however, are generally situated in a medical environment, surrounded by other health care professionals, and are held accountable for effective care delivery (Van de Bovenkamp et al., 2017), which may explain their relatively large focus on effectiveness.

The longlist of 46 QIs can be found in chapter 5 (Appendix 5-III). For each of the drafted QIs on the longlist, it is indicated whether it concerns a measure of structure (e.g. availability of facilities and means), process (e.g. medical interventions or interpersonal interactions) or outcome (e.g. improved quality of life as a result of health care processes or structures) (Blumenthal et al., 1996; Donabedian, 2005). Of the 46 drafted QIs, most address structures and processes of health care, only three address outcomes.

Furthermore, the drafted QIs apply to different organisational levels: individual level (quality of care as provided by one professional), the level of providers (quality of care as provided by hospitals, departments, teams or practices), and regional or national level (quality of care as provided in a region or entire country).

In addition, for each of the drafted QIs possible data collection methods are proposed. In order to obtain quality information on the different quality dimensions, different methods may be applied. Some information, for instance information on whether a certain test was performed, can be obtained by extracting data from electronic medical records (EMRs). This will probably not require extra registration, especially given the currently advanced data extraction techniques, or requires only simple registration such as checking a few boxes. Other information, for example experiences of a patient with provided health care or perceived health outcomes, may require more elaborated methods, such as questionnaires like patient reported experience measures (PREMs), patient reported outcome measures (PROMs), or other instruments such as observations or narrative methods. In chapter 4 it was agreed that measurement instruments should also be suitable for collecting information from people with DS themselves.

Preconditions and considerations for further development and use (Research question 3)

As indicated in chapter 5, the drafted longlist of QIs is not a ready-to-use indicator set. More work is needed by people with DS, their caregivers, (health care) professionals, health care managers, health insurers, the inspectorate, and other national, regional and/or local stakeholders, and researchers, in order to compose a QI-set, or perhaps QI-sets. This thesis identified considerations and preconditions to take into account for the further development and actual use of the QIs.

1. QI purposes

First of all, as mentioned above, the thesis identified the purpose(s) study participants want to achieve using the QIs: 1) to improve quality in health care by identifying potential areas for improvement and 2) to increase insight into available health care, enabling people with DS (and their caregivers) to make well-informed health care choices, and supporting health care professionals to make well-informed referrals (chapter 4). However, the two purposes for the QIs appear not to be easily compatible. Quality information provides insight into areas for improvement, which supports health care professionals to improve health care provision. In order for people with DS and their parents to make well-informed health care choices and find the right health care providers, this information should be publicly accessible. However, a finding of chapter 4 was that health care professionals working with people with DS are reluctant to make such information

publicly available and would rather keep this information only accessible to their own team for internal quality improvements (chapter 4). A similar attitude is seen among professionals working in care for people with Parkinson's disease (Damman et al., 2019). The participants in the Delphi-study (chapter 4) were concerned, perhaps as a result of extensive media attention to health care incidents in the past (Van de Bovenkamp et al., 2017), that publicly available information would lead to a 'shaming-and-blaming'-situation in which a safe working environment would be at risk, hampering health care quality. Thus, it may be difficult to achieve both purposes with the QIs, also because research has shown that although patients are encouraged to make well-informed health care choices, QIs seem to have little influence on these choices (Damman et al., 2019; Van de Bovenkamp et al., 2017; Victoor et al., 2016; Zwijnenberg et al., 2016). Moreover, the scarcity of well-trained, DS-specific, health care professionals, and for instance ID physicians, leaves little room for a free choice.

2. Large number of QIs

Secondly, the number of QIs and related registration burden of the users should be taken into account (chapter 4). The longlist of drafted QIs (chapter 5) contains 46 QIs, which is quite a large number, especially in the light of the registration burden already experienced by health care professionals (Blume et al., 2016) and anti-registration movements (Berwick et al., 2016; Ploegman et al., 2019). Let alone the efforts needed from people with DS and their caregivers to provide information (chapter 5). Registration burden may be diminished by allowing future users of the QIs to select a minimal dataset of the QIs for registration, or to alternate between QIs, registering some QIs one year and others the next (chapter 4). This could be facilitated by a modular structure of the QIs, enabling easy selection of QIs measuring quality items relevant to the user. Next to modules based on the content, specific modules, or QI-sets, could be developed per discipline in order to facilitate implementation (chapter 4). For example, a paediatrician provides different care and may be interested in different information than a primary care speech therapist. Whereas QIs on for example person-centredness may be the same for every discipline.

Perhaps the most effective measure for limiting registration burden, is to reduce the number of QIs by carefully selecting QIs for further development. Although all quality issues covered by the drafted QIs were considered relevant by the participants of the Delphi-study, the issues should be prioritised based on practical

and up-to-date considerations, in order to select QIs for further development. For example, the restrictive measures related to the COVID-19 pandemic, brought prevalent underlying ideas to light about whether and how health care should be provided (Auener et al., 2020). The need for some of the medical screenings was questioned and more telemedicine has been used (Bloem et al., 2020a). Such insights may be helpful in prioritising quality issues and the selection of most relevant QIs for further development (chapter 5). The newly developed Dutch guideline for health care for children with DS and the guideline for adults with DS which is currently under development (at the time of writing this Discussion) may be used to inform the selection of QIs. Moreover, for some of the identified quality issues, an obvious next step would rather be to develop other instruments, tools or interventions instead of, or next to, QIs. An example of drafted QIs that may be omitted (at least for now), are the ones about DS-specialised education of health care professionals. Such education could be an answer to the lack of DS-specific knowledge among health care professionals as perceived by parents of people with DS (chapter 3). However, it does not (yet) exist and should be developed first. Developing QIs measuring whether professionals have followed such education could be a second step (chapter 5).

3. Measuring structure, process, or outcome

A third point to consider relates to the type of QI (outcome, process or structure). Probably because of the considerable focus on person-centredness and organisation of care, a relatively large number of the drafted QIs address structures and processes of care (chapter 5). However, outcome indicators have long been considered as the preferable measure of health care quality (Porter, 2010), because outcomes are the product of processes and structures of care (Donabedian, 2005), and they are an indication of the quality of the processes and structures that have led to the outcomes. Also, for health care for people with chronic conditions and multimorbidity, such as people with DS, outcome measures are said to be important quality indicators, because they are able to provide an indication of the often multidisciplinary and complex care (Kourkoutas et al., 2010; Makovski et al., 2019). However, especially because of the complexity of multimorbidity care, it may not always be clear which processes or structures caused the outcome (Donabedian, 2005), and whether the outcome was even caused by health care at all. For example, an often used outcome measure is (health related) quality of life (Makovski et al., 2019), which is also present in the drafted QIs in this

thesis (chapter 5). A child with DS may score higher on quality of life because of better management of thyroid disease, but also because of better support at school. Outcome measurement instruments should thus be specific enough to distinguish outcomes that can be attributed to health care, and preferably to specific parts within health care. On the other hand, an advantage of a more general outcome measure, such as quality of life, is that it may result in a joint sense of responsibility, shared by all (health care) professionals involved. In addition, a problem that may occur when using more specific measures, is that standard specific measures are used, which overlook the unique values patients attribute to specific outcomes (Groenewoud et al., 2019; Wiering et al., 2016). For example, one person may consider pain reduction as most important, whereas another person may value functional ability much more. This is in line with our findings concerning the careful balance between burden and outcomes of treatment and the related quality sub-dimension "*Impact/burden of health care/treatment on patient's life and on his/her environment*". This balance can be different for each person and different for people with DS as compared to the general population (chapter 3). Measurement instruments should be sensible for such differences and the way in which outcomes are measured should be carefully considered and may encompass narrative or observational methods (Groenewoud et al., 2019). At the same time, since it will be difficult to take into account all such differences, one may also use process QIs measuring (perceived) involvement of people with DS and their caregivers in decisions and whether their values were respected. The above argues for a combination of outcome (specific and general), process and structure QIs, or perhaps a focus on process QIs, in order to provide an elaborated insight into health care for people with DS. This is corroborated by other research indicating that patients in long term care and with complex needs especially value process measures (Barelds et al., 2010; EXPH, 2019; Rademakers et al., 2011; RVS, 2020).

4. Data collection

Another consideration for the further development of QIs, already introduced in the above paragraph, is related to the instruments suitable for collecting the needed information. For some QIs, collecting information may require much more effort than for others and it may even be argued that as a first step, only QIs should be put into use that require the least amount of work. For example, if information is already being registered in electronic medical records (EMRs), efforts needed for

the collection of data are relatively small (chapter 5). However, its success depends on the accuracy of registration by professionals and the compatibility of different information systems used by different health care providers (Borousiak et al., 2018; Verheij et al., 2018). Instruments suitable for gathering information from people with DS would probably require more effort (chapter 2). The identified QIs for people with ID (chapter 2) could inform the development of such instruments. However, although instruments that are able to obtain information from people with DS would be meaningful, the use would almost inevitably place extra demands on people with DS and their caregivers. Moreover, an answer should be found to the question who will provide the information if the person with DS may not have the (total) capacity to provide it (chapter 4).

5. QIs as part of a learning system

The last and perhaps most important insight to take into account during further development and implementation of the drafted QIs relates to the application of an integrated care approach. In chapter 3 it was argued that the quality issues addressed by people with DS and their caregivers, call for an integrated care approach in which care is coordinated based upon the personal needs of patients (González-Ortiz et al., 2018). This is in line with the 'user-led definition' of integrated care, which is as follows: "My care is planned with people who work together to understand me and my carer(s), put me in control, coordinate and deliver services to achieve my best outcomes" (WHO Europe, 2016). The definition matches the findings in this thesis in many ways. First, the definition closely relates to the expressed need for collaboration between professionals and coordination of services (chapter 3). Second, the definition matches the identified importance of a right balance between benefits and burden of a disease or treatment (chapter 3), which is about deciding what are the "best outcomes". Third, in order to define the right balance, insight into a person's life (chapter 3), beyond the medical domain, is required (chapter 6). This relates to the part of "understand me and my carer(s)" and involves DS-specific communication and interaction (chapter 3). Furthermore, the "put me in control"-part of the definition of integrated care is reflected in our findings on the balance between a person's autonomy and values versus choosing the healthy option (chapter 6). Chapter 6 argues that this may be facilitated by collaborative decision-making approaches in which a person receives all needed support (human, material, or other) to make autonomous decisions (Peisah et al., 2013).

An integrated care approach is known to be especially important in health care for people with complex needs or chronic disease (Busetto et al., 2016; González-Ortiz et al., 2018), such as people with ID or DS (Buntinx & Schalock, 2010), and should therefore be the applied paradigm during further development and realisation of the drafted QIs. Literature on integrated care argues that many interventions, actions and initiatives may be needed to establish integration of care (Valentijn et al., 2013; WHO Europe, 2016). One of the elements of integrated care entails monitoring and evaluation in order to check and ensure quality of all these aspects (González-Ortiz et al., 2018; WHO Europe, 2016), which is where QIs come in. This implies that in order to improve health care for people with DS, QIs should not be introduced as a standalone improvement initiative, but preferably as part of a larger whole. This is in line with the model of 'learning health care systems' as mentioned in chapter 5. In learning health care systems, quality information ("summaries of performance against evidence-based standards") from different sites of the system is used in audits and feedback, in order to create value for patients, populations, providers and in terms of costs (Menear et al., 2019). The drafted QIs may provide this quality information in the learning health care system for people with DS. The fact that the QIs are formulated for different organisational levels is in line with such learning health care systems and principles of integrated care (Menear et al., 2019; Zonneveld et al., 2018). Additionally, chapter 5 also suggested other ways in which the identified quality dimensions could be used alongside QIs to improve health care for people with DS. For example, it was suggested to use the identified quality dimensions as input for evaluation meetings of health care professionals, or as topics for dialogues between a health care professional and the person with DS (and their caregivers). It was also suggested to use the quality dimensions as basis for a checklist that people with DS or caregivers could use to evaluate the received care or to structure the dialogue with health care professionals (chapter 5).

Impact of QIs on the lives of people with DS (Research question 4)

Since this thesis applied a bottom-up approach, the issues addressed by the drafted QIs matter to people with DS and their caregivers. Thus, assuming that the use of the drafted QIs leads to health care improvements, it can be expected that these improvements are meaningful to people with DS and, in turn, to better health and quality of life (Skotko et al., 2013). For example, the QIs could stimulate more person-centred care and collaborative approaches, in which people with DS, their caregivers, and health care professionals together discuss the needed

care and the way in which it is provided. This would contribute to a person's feeling of being seen and heard, which parents and support staff deemed very important in the lives of people with DS (chapter 3 & 6) and which is considered a contributor to quality of life (Schalock et al., 2005). The QIs could also increase awareness among health care professionals about the careful balance between burdens and gains of a certain treatment, and could motivate professionals to take into account the family or living context of people with DS (chapter 6). This would establish a good fit of the treatment into the lives of people with DS and better adherence (Rathert et al., 2012). However, (caregivers of) people with DS repeatedly stated that medical services were just one aspect of a larger total of services and supports needed for a person with DS to live his/her life (chapter 3 & 6). They also mentioned that they experienced struggles in applying for, coordinating, and aligning all needed services (chapter 6). Integration of all these services, within and outside of health care, for example with the help from a case manager or 'patient navigator' (Dimitropoulos et al., 2019), would be desirable to alleviate these struggles. This would contribute to the supportive environment that is needed to enable the lives of people with DS (chapter 6).

In addition, transparency of quality information, if QI scores are indeed published, can help (caregivers of) people with DS to find and choose the needed care. This may eliminate some of the difficulties related to finding the needed care and will contribute to a situation in which, caregivers experience less stress, people with DS receive suitable care, and health problems are treated. However, as mentioned before, it is not clear as to how and whether people with DS and their caregivers would actually use QIs to find the needed care (if available).

Strengths and limitations

Because of the relatively small amount of work that has been done in the field of quality of health care for people with DS (Kinnear et al., 2018; Kyrkou et al., 2018; Santoro et al., 2021), this thesis had an exploratory character and wide focus. This required (mainly) qualitative research methods (Tong et al., 2007), which allowed a thorough identification of all potential QIs from the perspective of people with DS (the 'patient perspective') and their caregivers as well as from the professionals' perspective. It also enabled an elaborate analysis of the preconditions for the

further development and implementation of the drafted QIs. Both important criteria when developing feasible and effective quality instruments (Kelley & Hurst, 2013; Kötter et al., 2013; Rathert et al., 2012; Santana et al., 2019; Wiering et al., 2017). Because of its wide focus, this thesis found differences in importance of quality issues as considered by health care professionals and by people with DS and their caregivers. This shows the added value of including the 'patient' perspective in quality improvement initiatives, which is still not always practiced (Kötter et al., 2013; Poitras et al, 2018; Rathert et al., 2013; Wiering et al., 2017) and ascertains that the drafted QIs cover all issues relevant in terms of both clinical relevance and meaningfulness for people with DS. However, because of the exploratory character of this thesis, the QIs are not 'ready-to-use'. They are formulated in a quite general manner, which makes them suitable for use in different health care disciplines, but perhaps not specific enough to measure discipline-specific quality aspects. Although much work is still to be done, this thesis provides a profound and firm basis for QIs for health care for people with DS.

This thesis is quite innovative because it actively involved people with DS. Although including people with ID in health research is increasingly popular (Frankena et al, 2015), studies specifically including people with DS are limited (chapter 2). During the interviews with people with DS, participants were supported to express their opinions, for example by using visuals, and by allowing them to invite a confidant to join the interview. However, interviews require a certain level of verbal skills, which are mostly only present in people with a mild to moderate level of ID (Bull, 2020; Patel et al., 2018). Additional ways to obtain information from people with DS, such as adding co-researchers with DS to the research team, or using more visuals or in a different way, would perhaps have enabled people with DS to express their opinions even better (Frankena et al., 2015; Zartler, 2014). However, interviews are often used and considered quite effective when it comes to involving people with ID in research (Frankena et al., 2015). The potential information gap, that may have existed in our findings, especially with regard to people with DS with more severe levels of ID, was probably filled by the interviews with parents and focus groups with support staff members and by inclusion of patient representative organisations in the Delphi-study (chapter 4).

The 'professional perspective' was covered by including a large variety of Dutch experts in the field of health care for people with DS within the Delphi-study

(chapter 4). The participating professionals represented almost all disciplines generally involved in health care for people with DS. On the one hand, the large diversity of participants has probably resulted in the large number of quality issues to be covered by the QIs. On the other hand, in this way it was possible to capture the large variety of quality issues experienced by the different disciplines. Moreover, it is argued that heterogeneity of participants in Delphi-studies contributes to widely accepted and credible QIs (Boulkedid et al., 2011).
Additionally, because of the heterogeneity of the research participants, reflecting the large variety of people with DS and their caregivers, and the multi-disciplines relevant in health care for people with DS, the findings are expected to be generalisable to all health care for people with DS in the Netherlands. The drafted QIs may also be applicable to other countries. However, especially the QIs addressing organisational issues of care may not (all) be applicable in other countries, as care may be organised differently there. Moreover, different (cultural) contexts may result in different choices for selection and further development.

The drafted QIs reflect quality issues that are similar to the ones mentioned in literature (IOM, 2001; WHO, 2018). This supports our findings, but it also shows that people with DS are not that different from the general population, in the sense that they have similar needs and preferences, which was not often studied before (chapter 2; Kinnear et al., 2018). It is even argued that quality of health care for people with DS may serve as indicator for quality of health care in general, because in health care for people with DS many issues are put under a microscope (Phelps et al., 2012). In that sense, quality of health care for people with DS can be considered the 'canary in the coal mine' for health care in general. Hence, our drafted QIs will not only be valuable in health care for people with DS - and ID, but for the health care system as a whole.

Next to this generalisability to the general population, the findings of this thesis may be specifically useful for health care for people with ID, since there are many similarities regarding topics like comorbidity, communication problems, and cognitive abilities (Bakker-van Gijssel et al., 2017; Kinnear et al., 2018). Moreover, the findings of this thesis may be informative for health care for all people with special needs and/or fragility. This may include older people, given the aging society and the increasing complexity of needs (Eriksson & Hellström, 2020; Tonelli et al., 2018), but also people with limited health literacy, such as migrants, partly

facing similar challenges as people with DS (Roodbeen et al., 2020). However, the thesis also illustrates that people with DS require different or more action in order to achieve the same level of fulfilment of needs as people without DS or people with ID (Kinnear et al., 2018; Phelps et al., 2012). For example, communication and interaction between a patient and a health care professional is considered important for the general population (WHO, 2018). However, it receives relatively strong attention in the drafted QIs, which is a reflection of the extra support that is required in order to meet the needs of people with DS, as compared to the general population.

Further reflection on findings

QIs in the Dutch health care system

This thesis provides a broad picture of quality of health care for people with DS that focuses on, but goes beyond, QIs. As was noted throughout the thesis, the multilevel and multidisciplinary character of health care for people with DS demands an integrated care approach (Buntinx & Schalock, 2010; Santana et al., 2018; Zonneveld et al., 2018), in order to create a learning health care system in which QIs provide information at different sites of the system for continuous learning and improvement (Menear et al., 2019). Such a systemic multilevel approach is needed in order for innovations to be successful and sustainable (Menear et al., 2019; Van den Driessen Mareeuw et al., 2015), especially since the current Dutch health care system is complex and consists of diverse coexisting quality initiatives and steering mechanisms ('layers') (Van de Bovenkamp et al., 2017). A 'layer' by which Dutch health care is predominantly governed is the market-based system, which was introduced in 2006. In this system with managed competition, health care insurers buy health care from health care providers who are supposed to compete on the basis of quality and price of the care they provide (De Vries et al., 2021; Van de Bovenkamp et al., 2017). In this system, health care providers compete for patients and health care insurers compete for the insured (Van de Bovenkamp et al., 2017). In this market-based health care system, insight into quality (and price) are very important, and stringent quality regulation and strict QIs, enforced by national authorities, are put in place (Van de Bovenkamp et al., 2017). As time passed, the system gradually became more and more subject to criticism, especially expressed by health care professionals, and it was questioned

whether it led to better and more affordable health care (De Vries et al., 2021). These sentiments resulted in more room and attention for local and informal improvement initiatives, often initiated by health care providers or professionals, and more focus on subjective quality measures and less on rigid QIs: another layer emerged (Van de Bovenkamp et al., 2017). Different layers may require different types or usages of QIs. While the market based system predominantly requires traditional QIs, informal improvement initiatives may use QIs more as quality criteria along which collaborative quality improvements could take place (Wells et al., 2018). Both types of QIs are present in the drafted QIs. The traditional ones are fore example QIs measuring the percentage of babies with DS who had their heart screening in time. The 'criteria' ones are for instance the drafted QIs about coordination or transition from child to adult health care. Research on measuring nursing care quality in hospitals shows a similar distinction between different types of QIs, based on different usages and users of quality information (Stalpers et al., 2016). Screening QIs ('traditional QIs') may be used by health insurers to monitor and compare hospitals, while hospitals or departments may prefer QIs measuring quality of care as perceived by professionals or patients, because such QIs provide information that can be used for internal quality improvements (Stalpers et al., 2016). The same research project also underlined that QIs are only worthwhile if they are used in an environment (e.g. a hospital or team) in which quality improvement is sufficiently incorporated in quality policies and working practices (Kieft et al., 2018). In line with the latter, the thesis also suggested to deploy other improvement initiatives alongside the QIs in order to 'really make a change'.

Advancing insights

The pluriformity of the drafted QIs and the fact that other improvement initiatives were also suggested in this thesis, is a result of advancing insights during the research project. During the course of the research project, it became clear that it was difficult to capture the complexity of health care for people with DS in straightforward 'traditional' QIs. This required a different, or additional, research paradigm that fitted with the complexity as identified by the findings of this thesis. Next to a paradigm in which accountability and improvement are key, also a paradigm based on flexibility and different perspectives had to be applied (Van Kemenade et al., 2021). It is argued that an integrated care approach requires application of different paradigms and interlinkages between them (Van

Kemenade et al., 2022). Van Kemenade et al. (2022) also argue that, in line with this thesis, a mixture of initiatives and actions is needed and that deploying QIs is one. They rather create a situation in which collaborative evaluation and learning take place during co-creation processes involving different actors in the system (Van Kemenade et al., 2022). QIs could inform such processes, along with other information based on experiences, context and values.

Future steps

Considering the above, it can be concluded that the drafted QIs should be introduced alongside other initiatives and as part of a learning health care system applying an integrated approach. The function of QIs would be to stimulate quality improvements at different sites in the system of health care for people with DS, thereby improving the total system and its coherence. In this way, introduction of the drafted QIs could actually induce, as Donabedian (2005) already argued, health care reforms.

Development of QIs: start with 'low-hanging fruit'

A next step for the development of QIs regardless of the system of which they may be part, is as follows. Given the complexity of health care for people with DS, it might be wise to start with the (relatively) 'low-hanging fruit': the information already registered in EMRs. An inventory should be made of what is already being registered by health care professionals who are providing care to people with DS. The newly developed guideline for adults with DS and the revised one for children could inform this process. The inventory could be started in Downteams, which are known to register information on children with DS. However, it should also be investigated whether primary care professionals, such as speech therapists, physiotherapists and dieticians, are registering information and which information it concerns. Professional organisations of the disciplines involved in health care for people with DS could play an important role in this inventory. Developing such specific QIs needs specific, probably less multidisciplinary work and could be one of the future steps for which this thesis forms a basis. For example, the drafted QIs do not cover the specific interventions or therapies a speech therapist or physiotherapist should use in people with DS. Furthermore, maybe less 'low hanging', is to investigate whether it would be possible to develop a simple

measurement instrument for people with DS and/or their caregivers. This process could for example be informed and supported by existing initiatives measuring person centred outcomes (www.platformuitkomstgerichtezorg.nl). If an inventory of EMR-registered data is made, and ideally a simple instrument for people with DS is developed, data scientists should be involved in order to set up methods for automated extraction of the available data from the different EMRs, and if possible of the simple measurement instrument for people with DS. This data extraction should result in comprehensible quality overviews ('dashboards') for the health care professionals involved (Mørkrid et al., 2021). This would provide health care professionals with information on their health care provision, on which they could base improvements.

However, a solution should still be found to the reluctance of health care professionals towards publicly available quality information, which may prevent people with DS and their caregivers from making well-informed health care choices. A Dutch network of professionals providing care to people with Parkinson's disease ('ParkinsonNet') seems to have found a solution (Bloem et al., 2020b). Professionals who are members of the network are obliged to take part in a quality monitor. Moreover, in order to become a member of the network, and as such be notified as specialist in Parkinson's disease (PD), professionals have to have a minimum of PD-patients under their supervision and they are obliged, among other things, to update their public (online) profile and take part in PD-specialised education and learning (ParkinsonNet, 2021). In this way, quality information is not made public, but only shared within ParkinsonNet. At the same time, health care professionals specialised in PD are visible to people with PD and to other health care professionals who can make referrals. Applied in health care for people with DS, such practice could be an acceptable compromise between on the one hand taking into account the reluctance among health care professionals to publish quality data, and on the other hand providing people with DS and their caregivers with sufficient information for making well-informed health care choices and finding the right health care providers. Although this may be helpful to people with DS and their caregivers, one could argue that more transparency by wider accessibility of quality data is desirable in order to ensure objective quality assessment. After all, if quality information would only be accessible for members within the network, the network may become introspective and less perceptive for indications of lower quality. Limited accessibility will also negate the advantages of open data sharing, such as more elaborated understandings of

health outcomes and insight into areas for improvements of health care provision (Kostkova et al., 2016).

A model for integrating services around the needs of people with DS

Establishing principles of learning systems and integrated care, requires, among others, joint efforts by all actors in the system, alignment of rules, flexibility, set up of collaborations, shared values, and sufficient means (Menear et al., 2019; Valentijn et al., 2013; Van den Driessen Mareeuw et al., 2015; Verheij, 2021). In addition, as the above shows, it may involve different paradigms, or at least the recognition of different paradigms (Van Kemenade et al., 2022). However, the efforts needed for establishing principles of learning systems and integrated care are worthwhile as they will result in better answers to the needs of people with DS. Such a systemic approach allows for integration of health care services with services outside health care, such as social care (Van Duijn et al., 2018). This may stimulate a more outward view among health care professionals, which goes beyond health care, and a more holistic approach towards people with DS. Below, it is sketched what several elements of such a systemic multilevel integrated approach could look like, what is already in place, and what steps are to be taken.

Regarding the needs of people with DS, this thesis has shown, in concordance with the literature, that these needs are complex and require a variety of services and supports to be answered (Kinnaer et al., 2018; Skotko et al., 2013). In response, so called 'Downteams' have been set up in the Netherlands (and elsewhere), including specialised health care professionals, who can be visited by a person with DS (and his caregivers) during one visit. Although Downteams are often seen as good practice, there are differences between the teams, and the composition of the team does not always match the needs of people with DS (Peters et al., 2020); for one person with DS not all disciplines may be relevant, whereas another person with DS may need even more disciplines than present in the team (chapter 3). Thus, answering personal needs also entails flexible 'mixing and matching' of health care services (Peters et al., 2020). This may involve primary care services or support at home, as well as highly specialised medical care. This may mean that the composition of Downteams, or other forms of regional collaboration initiatives, should be flexible. Additionally, it may require links with regional primary care services and support, but it may also entail the availability of a national, or perhaps larger-regional, team of experts who can be "flown-in" or consulted if needed.

Some of the Dutch Downteams already apply such flexible mixing and matching. A similar model is seen in the previously mentioned Parkinson case (Bloem et al., 2020b), as well as in an advice commissioned by the Dutch Ministry of Health on health care for people with rare conditions and complex needs who receive long term care (KPMG, 2019). Both network-based initiatives encompass national centres or collaboratives of expertise and local satellites. The national centres or collaboratives are responsible for keeping up-to-date with research and feeding the network with their expertise. They also keep on track with the satellites for identifying and collecting ideas to be used for research agenda setting (Bloem et al., 2020b; KPMG, 2019). The satellites, in collaboration with local health care professionals, provide, and ideally coordinate, general health care and support close to patients (Bloem et al., 2020b; KPMG, 2019). In health care for people with DS, these satellites could be the Downteams, which are ideally linked to local professionals such as, primary care physiotherapists, general practitioners, and social care. However, in the case of DS, despite some attempts, national (or supra-regional) centres or collaboratives of expertise are lacking. The existence of the centres of expertise, would enable Downteams to involve or consult extra expertise if this is required in order to meet the specific needs of a patient with DS. On the one hand, this would allow Downteams to flexibly mix and match the services based on the patient's needs and would secure accessibility of health care in the proximity. On the other hand, Downteams may choose to diminish the number of standard disciplines in the team, which may have a positive effect on health care costs.

Additionally, although Downteams are often mentioned as being well-coordinated care initiatives, they lack sufficient means to establish links with services outside health care (Peters et al., 2020). This thesis shows however, that coordination of *all* services is needed, including health care services and services outside health care. This calls for a 'patient navigator' (Dimitropoulos et al., 2019), who coordinates all services in the proximity of the person with DS. In the Parkinson model, such a 'personal care manager' operates at the intersection between the satellite and local services, including services outside health care (e.g. social services) (Bloem et al., 2020b).

Furthermore, as this thesis also showed, in order to identify the specific needs of people with DS, attention should be paid to the interaction and communication

between health care professionals and people with DS and their caregivers. Careful interaction and communication is important to be able to balance the benefits and burdens of a treatment and thus to define the best outcomes for the person with DS. Research shows that adapted communication and sensitivity to cues of patients may lead to better health outcomes (Di Blasi et al., 2001; Levinson et al., 2000; Schubbe et al., 2020). With regard to especially communication skills and conversation techniques it might be efficient to connect with research on, and initiatives for, people with ID in general, or other people with limited health literacy skills, such as migrants (Mastebroek et al., 2017; Roodbeen et al., 2020). However, ideally, such training may require specific DS-elements, because of the specific behavioural patterns and speech and information processing abilities related to DS (Bull, 2020; Grieco et al., 2015; Patel et al., 2018). Providing training to health care professionals in order to improve their communication strategies may be effective because health care professionals do not always seem to possess sufficient skills (Roodbeen et al., 2020). Additionally, research has shown that if extra effort is put in involving people with ID in their health decisions, their contributions are meaningful (Flynn et al., 2015). This calls for using shared or collaborative decision-making approaches (Peisah et al., 2013), which enables person-centred care that takes into account the person with DS, his values, abilities and preferences, and which not (only) focuses on his disease (Barry & Edgman-Levitan, 2012; Mittelmark et al., 2022; Santana et al., 2018). This also fits within the growing attention for, and increased considered importance of, person-centredness in health care (Santana et al., 2018). More focus on person-centredness may also more properly support people with DS in living their lives.

Conclusion and recommendations

To conclude, much effort may be needed in order to further develop and introduce the drafted QIs as part of a learning health care system, and to apply an integrated approach, including national expertise collaborations, local satellites, and patient navigators, in which attention is paid to adapted communication and person-centredness. These efforts are worthwhile because the proposed innovations lead to better answering the complex needs of people with DS, which would contribute to their lives, and may even result in lower costs (Bloem et al., 2020b). However, such innovations may not only benefit (health care for) people

with DS, but probably also the health care system as a whole, or at least health care for other people with ID, complex needs, or limited health literacy. In addition, such practices add to the growing body of knowledge on health care quality and integration. A first step could be to set up a working group consisting of medical experts, local and national policy makers, the Dutch DS association (representing (parents of) people with DS in the Netherlands), and researchers. This group should investigate possibilities and prerequisites for the formation of national expert collaboratives and the appointment of patient navigators. Also, information needs of people with DS and their caregivers should be further explored and answered, by QIs or for example by organisational changes. Furthermore, Downteams are recommended to identify data that are already registered in the EMRs they are using. They should be supported in this by data scientists and the hospital or other care providing organisation in which the Downteams are based. Doing all this requires an outward view, open mind, and readiness for acknowledging and applying different research paradigms, by all those who work with people with DS.

Nevertheless, in the meantime, this thesis ends with some low-key recommendations that may lead to perhaps tiny changes, but that may well be the start of larger innovations.

First of all, health care professionals are recommended to take time to carefully listen and look to the person with DS and caregiver(s) and try to obtain a real insight into his/her life, in order to be able to provide the care that matches the needs, preferences and values of this person and his/her context.

Second, health care professionals are urged to look further than their own discipline or their own working environment and to be open minded with regard to collaborations, with for example home support services.

Third, people with DS and their caregivers are recommended to provide insight into their lives during consultations or in other occasions, and to make sure that their needs, preferences, values and contexts are taken into account in health care (or other) decisions.

Last, all actors (potentially) involved in providing services and supports for people with DS, which in fact includes the whole society, are urged to actually see people with DS as part of our society, as part of our population, and to support them where they can.


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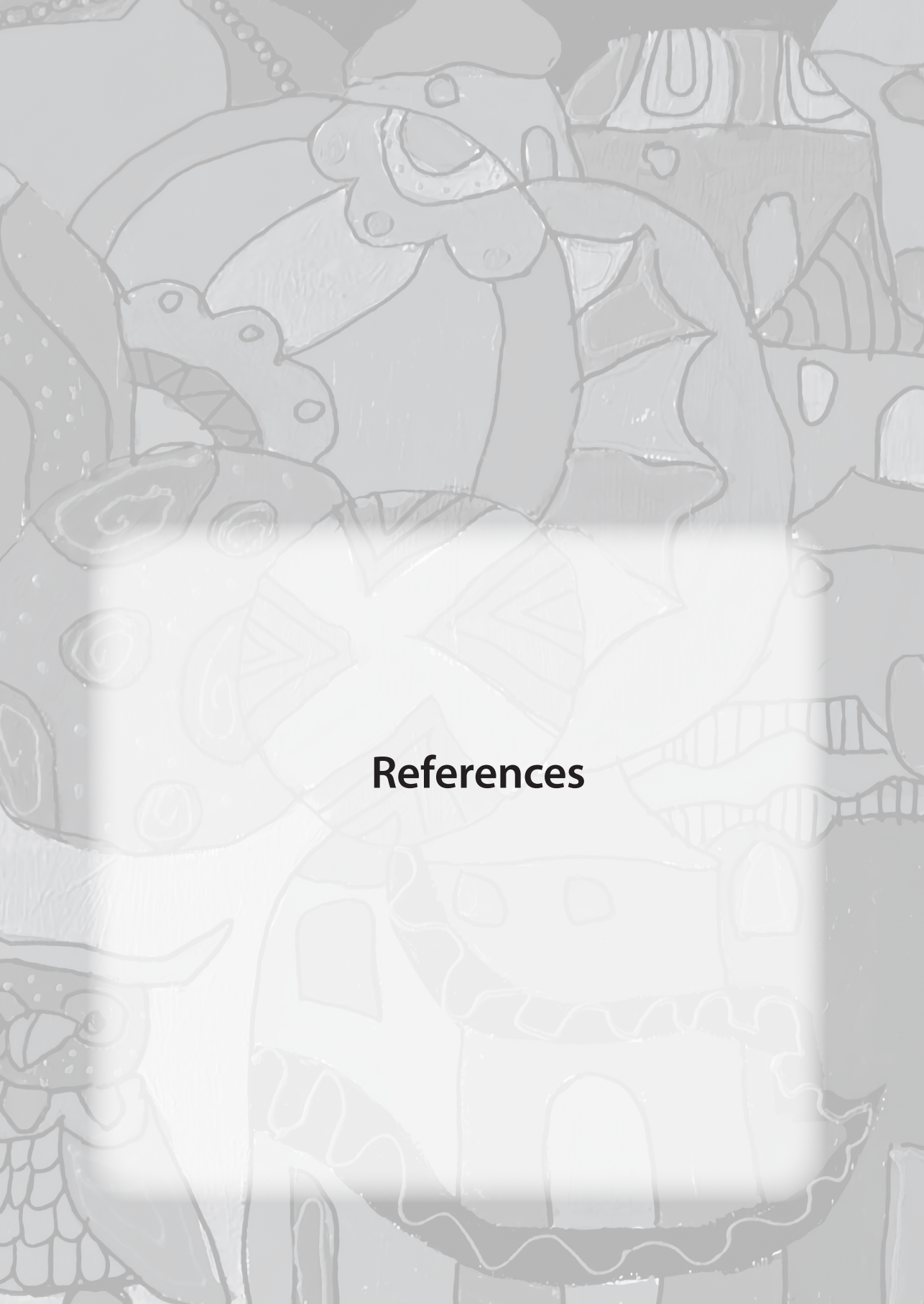

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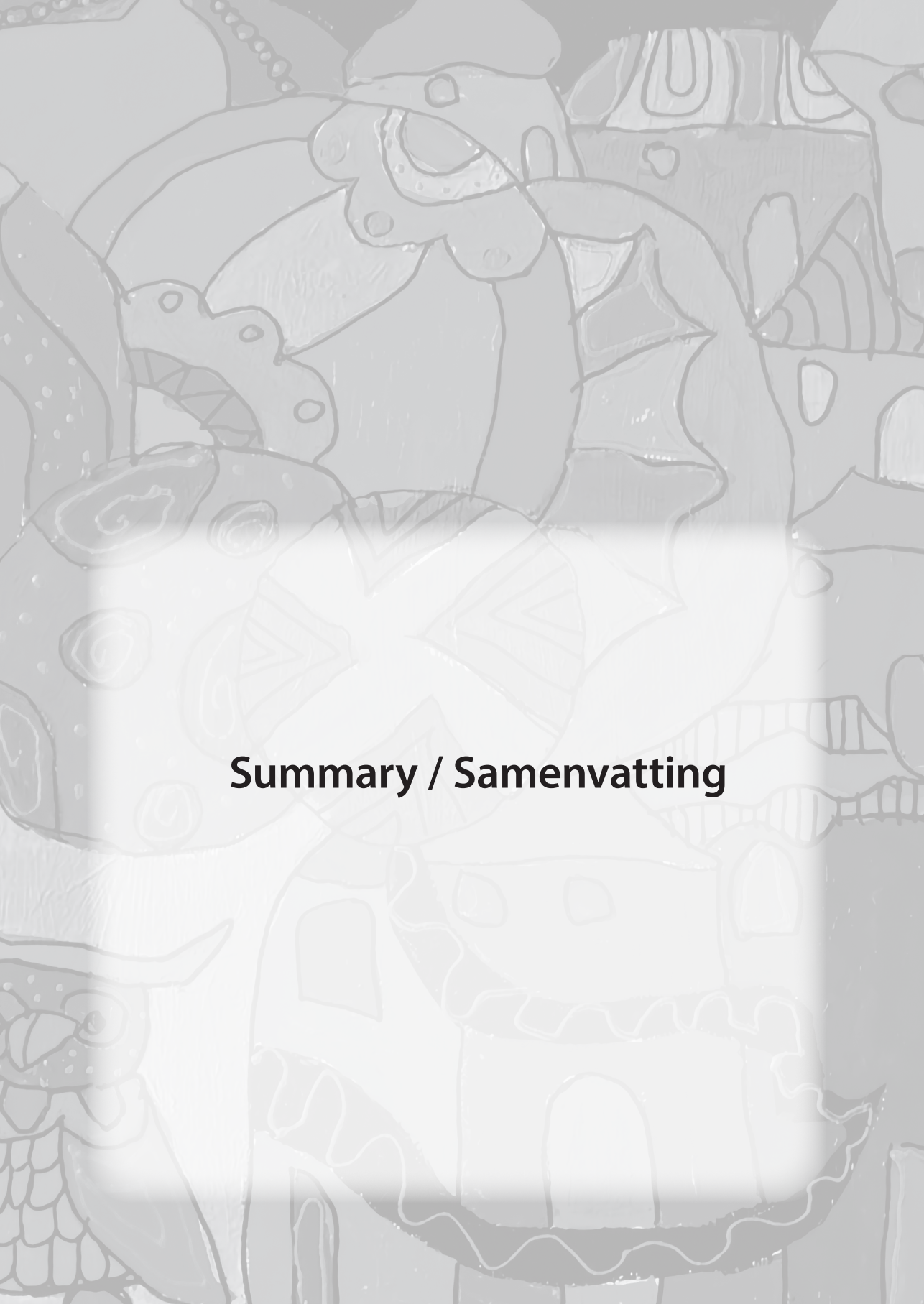

Summary / Samenvatting

SUMMARY

Background

Down syndrome (DS), or trisomy 21, is the most common cause of intellectual disability (ID) in humans and is related to a specific combination of physical and mental health problems. The number of people with DS in the Netherlands is about 12,700 (7.3 per 10.000 inhabitants). They form a substantial and relevant subgroup within the group of people with ID. This number, in combination with their specific needs, calls for dedicated means, services and policy. According to the Convention on the Rights of Persons with Disabilities (CRPD), people with disabilities, and thus people with DS, are entitled to all care and support required to live their lives according to their wishes and preferences. People with DS are reliant on a large variety of supports and services, one of which are health care services. Involved health care professionals may include a paediatrician or an ID physician, a speech therapist, a dietician, a physiotherapist, an ophthalmologist, an ear-nose-throat specialist and others, who may collaborate in multidisciplinary 'Downteams' in order to meet the complex needs of people with DS. It is acknowledged that because of their specific needs, high quality health care for people with DS is of vital importance. However, strikingly, little work has been done on quality of health care for people with DS and therefore their specific and complex needs are not always properly met. Quality indicators (QIs) are important instruments for quality improvement. QIs are measurable and carefully defined items of health care and provide insight into health care quality which in turn may identify directions for health care reforms, inform clinical decisions, and help patients finding the needed care.

It was the objective of this thesis to provide an empirically based first draft of such QIs and directions for their further development and use, as well as insight into the potential impact of QIs on the lives of people with DS.

Available QIs (chapter 2)

The first step of this dissertation was to investigate whether QIs for health care for people with DS did already exist. Therefore, a scoping review was conducted in search of such QIs, which is described in chapter 2. While conducting the review, using search terms for (synonyms of) Down syndrome, no QIs for health care for people with DS were found. Therefore, the search was extended to QIs for health care for people with ID, as these may also be useful for health care for people with

DS. The resulting 1478 hits were carefully screened and selected, which resulted in thirteen studies and thirteen indicators/indicator sets. One QI measured whether thyroid functioning in people with DS was checked. All other identified indicator sets were about health care for children or adults with ID. The settings to which the indicator sets applied differed largely, ranging from preventive or primary care to specific care chains or processes, and to national health systems. More than a third of the indicator sets focused on quality of supportive care and services. Often addressed topics were (multidisciplinary) collaboration, coordination and organisation of care and communication between care provider and person with ID. The QIs covering medical care primarily focused on screening and preventive care and barely addressed specific diseases and/or treatment courses. The quality of the indicator sets was evaluated using the AIRE-instrument (Appraisal of Indicators through Research and Evaluation), which showed that all sets had a clearly defined aim and setting description, had sufficiently involved relevant stakeholders in the development, and had provided supportive information or tools. There were large differences regarding the scientific evidence base of the sets. Most of the QIs in the indicator sets measured processes (e.g. measuring blood pressure) or outcomes (e.g. improved health) of care (about 40% each), whereas a smaller number of QIs measured structures of care (e.g. available resources) (about 20%). It was also investigated whether the six WHO quality domains (effectiveness, efficiency, accessibility, patient-centredness, equitability, safety) were covered by the sets. Effectiveness, efficiency, accessibility and patient-centredness were most addressed.

The identified scarcity of QIs for health care for people with DS justified the next elaborated steps of this dissertation towards such QIs. The found QIs informed those steps. Especially the QIs or indicator sets addressing (multidisciplinary) collaboration, coordination and organisation of care, and communication were useful, since these topics are not very DS-specific, but are important to all people with ID, including people with DS. In addition, the scoping review had stressed the importance of stakeholder involvement in QI development, which was a reason for involving relevant stakeholders in the following steps of the dissertation.

Empirical basis for the drafted QIs (chapters 3 and 4)

Chapter three describes a qualitative explorative study including semi-structured interviews with people with DS and with their parents and focus groups with support staff working in assisted living facilities for people with ID (including DS).

The study aimed to obtain insight into their perceptions regarding quality of health care for people with DS. This insight would form an important input for drafting the QIs. According to people with DS, it is important that health care professionals cure the health problem, communicate clearly, build a trust relationship, and also pay attention to other things in life that are not necessarily related to the health problem. Parents also underlined the importance of a holistic approach and added that coordination of all services involved, including services outside the medical domain, is an important element of health care quality. Support staff complemented that for people with DS respectful treatment and creating a feeling of 'being seen and heard' are also important elements of quality of health care. Parents and support staff indicated furthermore that the type of services/ professionals involved differs for each person with DS and that coordination of the transition from paediatric towards adult health care needs special attention.

The four-round Delphi study described in chapter four aimed to achieve consensus among health care professionals and patient organisations' representatives regarding the purposes, topics addressed and number of QIs to be developed. During this Delphi-study, the quality issues mentioned by people with DS, parents and support staff (chapter 3) were presented to the participants, as well as additional issues derived from the existing medical guideline for children with DS and issues regarding (development of) QIs for health care for people with DS. Participants could also propose additional issues. The participants agreed (consensus was achieved) that QIs for health care for people with DS should (have the purpose to): 1) Provide insight into available health care, enabling people with DS (and their caregivers) to make well-informed health care choices, and supporting health care professionals to make well-informed referrals; and 2) Provide information suitable for informing health care improvements. The participants stressed that QIs should not be used to judge health care professionals. Participants were concerned that QIs would make quality information about individual health care professionals publicly available, which would induce judgement of health care professionals and harm quality, instead of improving it. The study therefore concluded that patients' rights to relevant information have to be carefully balanced against providers' entitlement to a safe environment in which they can learn and improve. Furthermore, the participants opted for an evenly distributed mix of structure, process, and outcome QIs, covering the following quality issues: coordination and continuity of health care, effectiveness, safety, person-centeredness, and outcomes concerning health and quality of life. Additionally, participants argued

that the QIs should cover all health care disciplines involved in health care for people with DS. However, they urged to keep the number of QIs low, in order to prevent (administrative) burden for health care professionals and people with DS and/or caregivers. The participants had a tendency (but did not agree) to limit the coverage of the QI-set to the medical domain and exclude disciplines/services outside health care, such as support staff of assisted living facilities or family support. Furthermore, the participants noted that a QI-set should be tailored to different health care disciplines and information systems, and that instruments for collecting data should be suitable for people with DS. The participants also agreed that the development of QIs should be done with involvement of all relevant stakeholders.

Drafting the QIs (chapter 5)

In chapter 5 the quality issues proposed by people with DS, parents and support staff (chapter 3) and the quality issues agreed upon by health care professionals and patient organisations' representatives (chapter 4) were combined and clustered into groups, or sub-dimensions of quality. For each of the sub-dimensions, and based on the quality issues within the sub-dimension, QIs were drafted. The preliminary list of QIs was presented to relevant health care professionals and patient organisations, which resulted in refinements of the QIs and recommendations for the further development and use of the QIs. This finally led to a longlist of 46 potential QIs and 12 sub-dimensions, grouped into three main quality dimensions:

- Quality dimension 1: Effectiveness
  With sub-dimensions:
  - Timely recognition and adequate treatment of health problems
  - Expertise of providers
  - Safety
- Quality dimension 2: Organisation of care
  With subdimensions:
  - Organisation, coordination and continuity in general
  - Organisation, coordination and continuity specifically for Downteams
  - Organisation, coordination and continuity specifically related to transition from child to adult care

  - Accessibility: Health care nearby / within community or in primary care centres
- Quality dimension 3: Person-centredness
  With Sub-dimensions:
  - General
  - Impact/burden of health care/treatment on patient's life and on his/her environment
  - Involvement of all relevant stakeholders
  - Consideration of preferences and values of the person with DS and his/her family
  - Communication: Whether provider adapts communication to (dis)ability of patient and builds a trust relationship

The study also provided recommendations for selecting, further developing, and implementing QIs. First, the number of QIs should be limited. All relevant stakeholders should further prioritise and select the most relevant QIs for actual use. Current developments in health care could inform this prioritisation, such as anti-registration movements and changed insights regarding quality of health care. Second, high administrative burden should be prevented by facilitating easy data collection, for example by integrating the proposed QIs into existing electronic medical records (EMRs) and delineating existing instruments that might be suitable for collecting information from people with DS and/or their caregivers. Furthermore, a right balance should be found regarding transparency of quality information.

QIs in a wider perspective (chapter 6)

The qualitative exploration including interviews with people with DS and parents and focus groups with support staff, as described in chapter 3, did not only yield information on perceived (elements of) quality of health care. It also yielded information on the lives of people with DS and the potential impact of health care and QIs on their lives. It appeared that people with DS desire a life like others, a 'normal life'. The first group of findings provided insight into their leisure activities, their work or school, housing, and into barriers and levers for living a 'normal life'. The second group of findings related to participation in society, supporting services, the image of people with DS in media, and autonomy and its balance with making the healthy choice. The third group of findings showed the family perspective and addressed the efforts needed to arrange all the required support,

and the impact of having a child with DS. In the discussion, it was argued that health care professionals should apply a (more) person-centred approach, which would include a careful consideration of the elements of the life of a person with DS (as identified in this study), a more outward looking approach by health care professionals and collaborations with professionals from other disciplines or sectors, and shared-decision making practices. QIs could stimulate this and should thus reflect elements of such a person-centred approach.

Discussion

The research described in this thesis is the first studying QIs for health care for people with DS in such a thorough way, including people with DS, their caregivers and health care providers. Because of the relatively small amount of work that has been done in this field, this thesis had an exploratory character and wide focus, which resulted in a large number of drafted QIs. Therefore, careful prioritising and selecting the drafted QIs for further development is required, which is preferably done by all relevant stakeholders. By doing this, considerations regarding registration burden, the type (structure, process, outcome) of QIs, and data collection methods should be taken into account. It should for example be investigated which quality information is already being registered in EMRs and which additional information is still to be collected. Additionally, the extent to which quality information is accessible should be considered. Furthermore, in the discussion it is argued that, because of the multidisciplinary and complex character of health care for people with DS, QIs should be developed and introduced as part of a learning health care system, in which QIs provide insight into, and stimulate, quality at different sites in the system. Such a system preferably also includes other quality improvement initiatives, such as education activities and setting up collaborations, and requires an environment in which quality improvement is sufficiently incorporated in quality policies and working practices. Principles of integrated care are suggested for achieving this, as these encompass a systemic approach based on the needs of the person. This would lead to a better fit between provided health care and personal situation of the person with DS and better health outcomes, which would contribute to quality of life. A network-based model including national centres of DS-expertise, regional satellites, and personal coordinators, is proposed to put principles of integrated care into practice. Lastly, the discussion calls for a more supportive environment in which people with DS are truly part of the society.

SAMENVATTING

Inleiding

Downsyndroom (DS), of trisomie 21, is de meest voorkomende genetische oorzaak voor een verstandelijke beperking (VB) bij mensen en gaat gepaard met een specifieke combinatie van lichamelijke en geestelijke gezondheidsproblemen. Het aantal mensen met DS in Nederland is ongeveer 12 700 (7,3 per 10 000 inwoners). Ze vormen een substantiële en relevante subgroep binnen de groep van mensen met een VB. De omvang van de groep mensen met DS en hun specifieke behoeften, vragen om specifieke middelen, voorzieningen en beleid. Volgens het Verdrag voor de rechten van mensen met een handicap, hebben mensen met beperkingen, en dus mensen met DS, recht op alle zorg en ondersteuning die zij nodig hebben om hun leven volgens hun eigen voorkeuren in te richten. Voor mensen met DS geldt dat zij veelal afhankelijk zijn van verschillende soorten zorg en ondersteuning, waarvan medische zorg er één is. Bij de medische zorg zijn vaak een kinderarts of arts voor verstandelijk gehandicapten (arts VG), een logopedist, een diëtist, een fysiotherapeut, een oogarts, een KNO-arts en anderen betrokken. Ook zijn er multidisciplinaire teams, 'Downteams' of 'Downpoli's', waarin deze en/of andere professionals samenwerken. Deze teams zijn opgezet om aan de veelal complexe behoeften van mensen met DS te kunnen beantwoorden.

Door hun specifieke behoeften, is het voor mensen met DS extra belangrijk dat de zorg die zij ontvangen van goede kwaliteit is. Het blijkt echter dat er nog veel gedaan kan worden om deze kwaliteit te waarborgen en om ervoor te zorgen dat er aan hun complexe zorgvragen tegemoet wordt gekomen. Kwaliteitsindicatoren kunnen een belangrijke bijdrage leveren aan het verbeteren en waarborgen van kwaliteit. Kwaliteitsindicatoren zijn meetbare en zorgvuldig geformuleerde zorgelementen die inzicht kunnen geven in kwaliteit van zorg. Ze kunnen mogelijkheden voor verbetering in kaart brengen, als basis dienen voor medische beslissingen en patiënten informatie bieden bij het maken van een keuze voor de best passende zorgaanbieder.

Dit proefschrift had als doel om op basis van empirisch onderzoek een eerste aanzet te geven voor dergelijke kwaliteitsindicatoren en om richting te geven aan de verdere ontwikkeling en het toekomstige gebruik van de indicatoren. Daarnaast werd een beeld geschetst van de mogelijke impact van dergelijke indicatoren op de levens van mensen met DS.

Zijn er al kwaliteitsindicatoren beschikbaar? (hoofdstuk 2)

De eerste stap van dit proefschrift was om in kaart te brengen of kwaliteitsindicatoren voor de zorg voor mensen met DS al bestonden. Dit gebeurde met behulp van literatuuronderzoek dat is beschreven in hoofdstuk 2 (een zogenaamde 'scoping study'). Dit literatuuronderzoek, dat zoektermen voor (synoniemen voor) downsyndroom gebruikte, resulteerde in geen enkele kwaliteitsindicator voor de zorg voor mensen met DS. Daarom werd de zoekstrategie uitgebreid naar kwaliteitsindicatoren voor alle mensen met een VB. Het idee hierachter was dat deze ook bruikbaar zouden kunnen zijn voor de specifieke zorg voor mensen met DS. Uit de 1478 artikelen die de zoektocht opleverde, werden na zorgvuldige screening en selectie uiteindelijk dertien studies geïncludeerd en ook dertien kwaliteitsindicatoren of indicatorensets. Eén van de gevonden kwaliteitsindicatoren werd gebruikt om te controleren of de schildklierfunctie van mensen met DS regelmatig werd gecontroleerd. Alle andere indicatoren hadden betrekking op mensen met een VB. De settingen waarop de gevonden indicatoren van toepassing waren, liepen uiteen van preventieve of eerstelijnszorg tot specifieke zorgprocessen en nationale zorgsystemen. Onderwerpen die in de gevonden indicatoren vaak terugkwamen waren (multidisciplinaire) samenwerking, coördinatie en organisatie van zorg en communicatie tussen zorgverleners en mensen met een VB. De meer medisch georiënteerde indicatoren, gingen vooral over screening en preventie. Specifieke aandoeningen of behandelingen kwamen nauwelijks aan bod. De kwaliteit van de gevonden kwaliteitsindicatoren(sets) werd beoordeeld met behulp van het AIRE-instrument (Appraisal of Indicators through Research and Evaluation (Beoordeling van indicatoren door onderzoek en evaluatie)). Uit deze beoordeling werd duidelijk dat alle indicatoren een duidelijk gedefinieerd doel hadden, een duidelijke beschrijving van de setting bevatten, ontwikkeld waren met voldoende relevante stakeholders en dat er ondersteunend materiaal beschikbaar was. Er waren grote verschillen tussen de indicatoren wat betreft hun wetenschappelijke onderbouwing. De meeste indicatoren hadden betrekking op processen van zorg (zoals het meten van bloeddruk) of op uitkomsten (zoals verbeterde gezondheid). Het aandeel van zowel proces- als uitkomstindicatoren was ongeveer 40% (40% elk), terwijl ongeveer 20% van de gevonden indicatoren betrekking had op structuur van zorg (zoals beschikbaar personeel). Er werd ook in kaart gebracht in hoeverre de zes kwaliteitsdomeinen (effectiviteit, efficiëntie, toegankelijkheid, patiëntgerichtheid, gelijkheid en veiligheid) van de Wereld Gezondheidsorganisatie (WHO) terugkwamen in de

gevonden kwaliteitsindicatoren. Effectiviteit, efficiëntie, toegankelijkheid en patiëntgerichtheid kwamen het vaakst terug in de gevonden indicatoren.
Het minieme aantal gevonden indicatoren voor de zorg voor mensen met DS maakte duidelijk dat er voor het ontwikkelen van dergelijke indicatoren nog uitgebreid onderzoek nodig was: het onderzoek dat in de volgende hoofdstukken van dit proefschrift beschreven is. De gevonden kwaliteitsindicatoren voor mensen met een VB werden wel gebruikt als input voor dit vervolgonderzoek. Vooral de kwaliteitsindicatoren over samenwerking, coördinatie en organisatie van zorg en over communicatie waren bruikbaar, omdat deze onderwerpen niet erg DS-specifiek zijn, maar voor álle mensen met een VB relevant zijn. Bovendien had het literatuuronderzoek laten zien dat het belangrijk is om alle relevante stakeholders bij de ontwikkeling van kwaliteitsindicatoren te betrekken. Dit was dan ook een reden om deze stakeholders in de vervolgstappen van het promotieonderzoek te betrekken.

Empirische basis voor de ontwikkeling van de kwaliteitsindicatoren (hoofdstukken 3 en 4)

Hoofdstuk drie beschrijft een kwalitatieve exploratieve studie waarbij semigestructureerd interviews met mensen met DS en met hun ouders werden gehouden en focusgroepen met (persoonlijk) (woon)begeleiders van mensen met DS die in een woonvoorziening wonen. Deze studie had als doel om inzicht te verkrijgen in hun percepties aangaande kwaliteit van zorg voor mensen met DS. Dit inzicht diende als belangrijke input voor de op te stellen kwaliteitsindicatoren. Volgens mensen met DS is het belangrijk dat zorgprofessionals het gezondheidsprobleem verhelpen, dat ze duidelijk communiceren, dat ze ook aandacht hebben voor andere dan medische of gezondheids-gerelateerde zaken en dat er een vertrouwensband wordt opgebouwd. Ouders noemden ook het belang van een holistische benadering door zorgprofessionals en gaven aan dat coördinatie van alle zorg en ondersteuning, inclusief niet-medische zorg en ondersteuning, in hun ogen van wezenlijk belang is voor kwalitatief hoogwaardige zorg. Begeleiders voegden daaraan toe dat het voor mensen met DS ook belangrijk is dat er sprake is van respectvolle behandeling door zorgprofessionals en dat mensen met DS het gevoel krijgen dat zij gezien en gehoord worden. Ouders en begeleiders gaven verder aan dat elke persoon met DS andere (gepersonaliseerde) zorg en ondersteuning nodig heeft en dat de transitie van kinder- naar volwassenenzorg bijzondere aandacht verdient.

De Delphistudie uit hoofdstuk vier bestond uit vier ronden en had als doel om consensus onder zorgprofessionals en enkele vertegenwoordigers van patiëntenorganisaties te bewerkstelligen over gebruiksdoelen, onderwerpen en aantal van de op te stellen kwaliteitsindicatoren. Aan de deelnemers van deze Delphistudie werden de elementen van kwaliteit van zorg voorgelegd die door mensen met DS, ouders en begeleiders genoemd waren (hoofdstuk drie). Andere onderwerpen die werden voorgelegd, betroffen aanbevelingen uit de medische richtlijn voor de kinderen met DS en onderwerpen die te maken hadden met (het ontwikkelen en gebruiken van) kwaliteitsindicatoren. De deelnemers konden ook onderwerpen toevoegen die zij relevant achtten. De deelnemers werden het erover eens (er was consensus) dat kwaliteitsindicatoren voor de zorg voor mensen met DS twee gebruikersdoelen zou moeten hebben: 1) inzicht geven in beschikbare zorg, op basis waarvan mensen met DS en hun verzorgers hun keuzes voor passende zorg kunnen maken, en op basis waarvan zorgprofessionals kunnen verwijzen; 2) inzicht geven in verbetermogelijkheden. De deelnemers benadrukten dat kwaliteitsindicatoren niet gebruikt zouden moeten worden om zorgprofessionals op af te rekenen. Ze waarschuwden dat een situatie waarin kwaliteitsinformatie over individuele zorgprofessionals openbaar is, ertoe zou kunnen leiden dat zorgprofessionals publiekelijk zouden worden beoordeeld op hun functioneren, wat de kwaliteit van de zorg niet ten goede zou komen. Een conclusie van hoofdstuk vier is dan ook dat er een juiste balans moet zijn tussen het recht van de patiënt op relevante informatie aan de ene kant, en een veilige werkomgeving voor zorgprofessionals waarin zij kunnen leren en verbeteren aan de andere kant. Verder vonden de deelnemers dat de kwaliteitsindicatoren moesten bestaan uit een gelijke verdeling van uitkomst-, proces- en structuurindicatoren. Daarnaast zouden volgens de Delphi-deelnemers de volgende onderwerpen in de kwaliteitsindicatoren aan bod moeten komen: coördinatie en continuïteit van zorg, effectiviteit, veiligheid, persoonsgerichtheid en uitkomsten met betrekking tot gezondheid en kwaliteit van leven. Ook zouden de kwaliteitsindicatoren bruikbaar moeten zijn voor alle zorgprofessionals die betrokken zijn bij de zorg voor mensen met DS. Men vond echter ook dat het aantal kwaliteitsindicatoren zo klein mogelijk zou moeten zijn, om de administratieve last voor zorgprofessionals en voor mensen met DS en hun verzorgers laag te houden. De deelnemers neigden er ook naar (maar er was geen consensus) om in de kwaliteitsindicatoren alleen medische thema's en disciplines mee te nemen en niet-medische onderwerpen, zoals begeleiding of dagbesteding, eruit te laten. De deelnemers

vonden verder dat de kwaliteitsindicatoren geschikt zouden moeten zijn voor verschillende disciplines en informatiesystemen, en dat dataverzameling zou moeten gebeuren met instrumenten die geschikt zijn om informatie van mensen met DS zelf te achterhalen. Ten slotte waren deelnemers het erover eens dat de ontwikkeling van kwaliteitsindicatoren samen met alle relevante stakeholders plaats zou moeten vinden.

Een eerste concept van kwaliteitsindicatoren (hoofdstuk 5)

De kwaliteitsonderwerpen genoemd door mensen met DS, ouders en begeleiders (hoofdstuk 3) en de onderwerpen waarover consensus was onder zorgprofessionals en vertegenwoordigers van patiëntenorganisaties (hoofdstuk 4) werden in hoofdstuk 5 samengebracht en gecategoriseerd in groepen, of sub-dimensies van kwaliteit. Gebaseerd op de kwaliteitsonderwerpen in de betreffende sub-dimensie, werden voor elke sub-dimensie één of meerdere kwaliteitsindicatoren opgesteld. Dit leidde tot een voorlopige lijst met kwaliteitsindicatoren die aan relevante zorgprofessionals en patiëntenorganisatie werd voorgelegd. Op basis van hun opmerkingen werden de kwaliteitsindicatoren aangescherpt en werden aanbevelingen voor de verdere ontwikkeling geformuleerd. Uiteindelijk is een longlist van 46 mogelijke kwaliteitsindicatoren opgesteld, verdeeld over drie kwaliteitsdomeinen en in totaal twaalf sub-dimensies:

- Kwaliteitsdimensie 1: Effectiviteit
  Met sub-dimensies:
  - o Tijdige herkenning en adequate behandeling van gezondheidsproblemen
  - o Expertise van zorgverleners
  - o Veiligheid
- Kwaliteitsdimensie 2: Organisatie van zorg
  Met sub-dimensies:
  - o Organisatie, coördinatie en continuïteit van zorg (algemeen)
  - o Organisatie, coördinatie en continuïteit van zorg bij Downteams
  - o Organisatie, coördinatie en continuïteit van zorg met betrekking tot de transitie van kinder- naar volwassenenzorg.
  - o Toegankelijkheid: Zorg in de buurt of in eerstelijns gezondheidscentra
  - Kwaliteitsdimensie 3: Persoonsgerichtheid

Met sub-dimensies:

- Algemeen (passen zorgverleners een persoonsgerichte benadering toe?)
- Positieve of negatieve invloed van zorg of behandeling op het leven van patiënten en zijn/haar omgeving
- Betrekken van alle relevante stakeholders
- Het meenemen van voorkeuren en persoonlijke waarden van de persoon met DS en diens familie
- Communicatie: Of communicatie van de professional aangepast is aan de mogelijkheden en beperkingen van de patiënt en of er sprake is van een vertrouwensband.

Naast de longlist van de kwaliteitsindicatoren formuleerde de studie ook aanbevelingen voor hun selectie, verdere ontwikkeling en implementatie. Allereerst zou het aantal kwaliteitsindicatoren zo laag mogelijk moeten zijn. Alle relevante stakeholders zouden de kwaliteitsindicatoren moeten prioriteren en bepalen welke het meest relevant en geschikt zijn voor daadwerkelijk gebruik. Bij deze prioritering zouden huidige ontwikkelingen in de zorg meegenomen kunnen worden. Er kan hierbij gedacht worden aan initiatieven als 'Ontregel de zorg' en veranderde inzichten over kwaliteit van zorg. Daarnaast zou de administratieve last voor zowel professionals als mensen met DS en hun verzorgers zo laag mogelijk moeten zijn, bijvoorbeeld door de voorgestelde kwaliteitsindicatoren te integreren in bestaande elektronische patiënten-of cliëntendossiers (EPDs/ECDs), en door bestaande instrumenten in kaart te brengen en/of aan te passen die geschikt zijn om informatie te verzamelen onder mensen met DS en hun verzorgers. Verder zou er aan de ene kant gestreefd moeten worden naar transparantie van kwaliteitsinformatie, terwijl aan de andere kant voorkomen moet worden dat individuele zorgverleners publiekelijk worden afgerekend op de geleverde zorg.

Kwaliteitsindicatoren in breder perspectief (hoofdstuk 6)

De interviews met mensen met DS en ouders en de focusgroepen met begeleiders, zoals beschreven in hoofdstuk 3, leverde niet alleen informatie op over (ervaren) kwaliteit van zorg, maar ook over het leven van mensen met DS en de mogelijke invloed van kwaliteitsindicatoren daarop. Zo bleek dat mensen met DS het liefst een 'normaal leven' willen leiden, niet anders dan andere mensen. De eerste groep bevindingen gaf inzicht in hun vrije tijdsbesteding, hun werk of opleiding, woonomstandigheden en in bevorderende en belemmerende factoren

voor een 'normaal leven'. De tweede groep bevindingen beschreef participatie in de maatschappij, ondersteuning, het beeld van mensen met DS in de media en de autonomie van mensen met DS (o.a. bij het maken van gezondheidskeuzes). De derde groep bevindingen belichtte het perspectief van de familie, het regelen van alle benodigde ondersteuning en de invloed van een kind met DS op het gezin. In de discussie van dit hoofdstuk werden zorgprofessionals opgeroepen om een (meer) persoonsgerichte benadering te gebruiken, waarbij alle elementen van het leven van een persoon met DS (zoals in dit hoofdstuk beschreven) nadrukkelijk meegenomen worden, waarbij zij verder kijken dan hun eigen discipline of werkomgeving en open staan voor samenwerking met andere disciplines en sectoren, en waarbij gedeelde besluitvorming plaatsvindt. Kwaliteitsindicatoren kunnen een dergelijke benadering stimuleren en zouden daarom ook elementen van persoonsgerichte zorg moeten bevatten.

Discussie

Het onderzoek beschreven in dit proefschrift is het eerste dat de ontwikkeling van kwaliteitsindicatoren voor de zorg voor mensen met DS op een grondige manier heeft bestudeerd en daarbij mensen met DS, ouders, begeleiders en zorgprofessionals heeft betrokken. Doordat er op dit gebied nog relatief weinig onderzoek is gedaan, heeft het onderzoek beschreven in dit proefschrift een verkennend karakter en een brede focus, wat heeft geleid tot een groot aantal concept-kwaliteitsindicatoren. De kwaliteitsindicatoren dienen dan ook zorgvuldig geprioriteerd en geselecteerd worden, idealiter door alle relevante stakeholders. Hierbij zouden zaken als administratieve last, het type indicator (structuur-, proces-, uitkomst-) en instrumenten om informatie te verzamelen, meegenomen moeten worden. Er zou bijvoorbeeld uitgezocht moeten worden welke kwaliteitsinformatie reeds in EPDs/ECDs geregistreerd wordt en welke informatie nog mist. Ook moet bepaald worden in welke mate kwaliteitsinformatie toegankelijk is en voor wie. De discussie beschrijft daarnaast dat door het multidisciplinaire en complexe karakter van de zorg voor mensen met DS, kwaliteitsindicatoren ontwikkeld en geïmplementeerd moeten worden als onderdeel van een groter geheel, een lerend systeem waarin kwaliteitsindicatoren op verschillende plekken in dat systeem inzicht geven in de kwaliteit en daarbij kwaliteitsverbetering stimuleren. Een dergelijk systeem bevat idealiter ook andere manieren van kwaliteitsbevordering, zoals onderwijsactiviteiten en het opzetten van samenwerkingsverbanden. Hiervoor dient kwaliteitsverbetering voldoende

geïncorporeerd te zijn in (kwaliteits)beleid en in het dagelijks werk. In de discussie wordt de suggestie gedaan om hiertoe een geïntegreerde zorgaanpak te kiezen, omdat geïntegreerde zorg een systemische benadering toepast en daarbij de cliënt/patiënt centraal stelt. De gedachte is dat dit leidt tot een betere aansluiting van de zorgverlening op de persoonlijke situatie van de persoon met DS en tot betere gezondheidsuitkomsten, wat uiteindelijk bijdraagt aan de kwaliteit van leven. Er wordt voorgesteld om de geïntegreerde zorgbenadering in praktijk te brengen door een netwerkmodel te gebruiken dat bestaat uit nationale expertisecentra voor DS, regionale satellieten (bijvoorbeeld de Downteams) en persoonlijke coördinatoren. Ten slotte wordt in de discussie opgeroepen te zorgen voor een omgeving waarin mensen met DS goed ondersteund worden, zodat zij werkelijk deel uit kunnen maken van de maatschappij.


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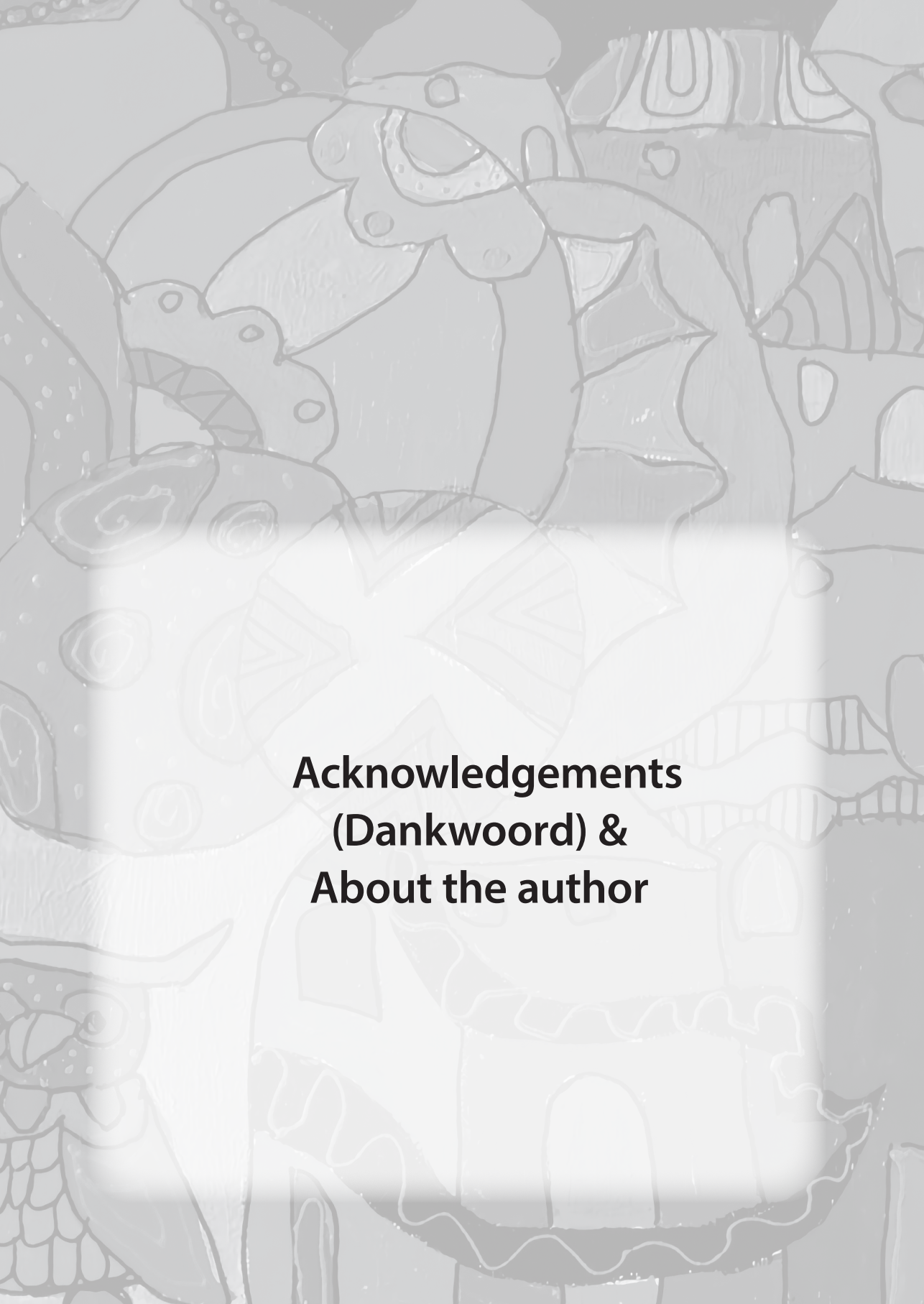

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About the author

Curriculum Vitae (in English)

Francine van den Driessen Mareeuw was born in The Hague (The Netherlands) on the 23rd of August, 1982. After completing her secondary (pre-university) education at the 'Haags Montessori Lyceum' in The Hague, she studied Nutrition and Health (BSc and MSc) at Wageningen University. She wrote her master thesis about the knowledge base of a Dutch programme aiming to promote physical activity among youngsters. She also did an internship at Protéines, a French health consultancy agency in Paris (France), where she scientifically and practically contributed to expanding a French programme aiming to prevent childhood obesity to other European countries. After completing this two year master programme (specialisation Public Health), she worked as a junior researcher at the Health and Society Group of Wageningen University on a project about knowledge exchange within the public health sector. There, she was offered the chance to participate in a summer school on health promotion (salutogenesis) by the European Training Consortium in Public Health and Health Promotion, which took place at the university of Cagliari, Sardinia (Italy). When the public health knowledge exchange project in Wageningen had finished, Francine was involved in several short term (research) activities, at 'STAP' (institute specialised in alcohol policy), TNO (research organisation), and other organisations. Then she returned to academia as a research assistant at the department of primary and community care of the Radboud University Medical Centre in Nijmegen. There she worked on different research projects related to (international) migrant health, work-related health and health of people with intellectual disabilities. The latter provided an excellent basis for the PhD-position on quality indicators for health care for people with Down syndrome, which she started in 2015 and which resulted in this PhD-thesis. The Jeroen Bosch Hospital in Den Bosch financially enabled this PhD-position at Tranzo, Scientific Centre for Care and Wellbeing, Tilburg School of Social and Behavioral Sciences, Tilburg University. As of May 2020, Francine works at SKILZ, an organisation developing scientifically and practically based guidelines for professionals working in (health) care for people with intellectual disabilities and older people. Francine likes nature and outdoor activities and is passionate about performing (singing) and listening to (classical) music. She lives with her partner Pieter and her sons Hugo, Philip and Otto in Arnhem.

Curriculum Vitae (in het Nederlands)

Francine van den Driessen Mareeuw is op 23 augustus 1982 geboren in Den Haag. Daar behaalde ze haar gymnasiumdiploma aan het Haags Montessori Lyceum, waarna ze Voeding en Gezondheid ging studeren aan Wageningen Universiteit. Na het behalen van haar bachelorsdiploma startte ze met de twee jaar durende master Voeding en Gezondheid (specialisatie Public health). Haar masterscriptie ging over de kennis die ten grondslag lag aan een Nederlands beweegprogramma voor leerlingen van het VMBO. Ook deed ze als onderdeel van haar master een stage bij Protéines, een adviesbureau op het vlak van gezondheid, gevestigd in Parijs (Frankrijk). Daar werkte ze aan een Frans programma gericht op de preventie van obesitas bij kinderen en droeg op wetenschappelijke en praktische manier bij aan de uitbreiding van dat programma naar andere Europese landen. Na het behalen van haar masterdiploma, ging ze als junior onderzoeker werken bij de Leerstoelgroep Gezondheid en Maatschappij van Wageningen Universiteit. Ze werkte daar aan een project over kennisuitwisseling binnen de publieke gezondheidszorg. Ook kreeg ze daar de kans om deel te nemen aan een summer school over gezondheidsbevordering (salutogenese) aan de universiteit van Cagliari (Sardinië, Italië), georganiseerd door het European Training Consortium in Public Health and Health Promotion. Toen het project over kennisuitwisseling in Wageningen afgerond was, was Francine betrokken bij verschillende kortdurende (onderzoeks)activiteiten, waaronder bij STAP (instituut voor alcoholbeleid) en TNO. Vervolgens keerde ze terug naar de wetenschap als onderzoeksmedewerker bij de afdeling Eerstelijnsgeneeskunde van het RadboudUMC in Nijmegen. Daar werkte ze aan nationale en internationale projecten over o.a. de zorg voor migranten, arbeid en gezondheid en de zorg voor mensen met een verstandelijke beperking. Met name de projecten over het laatste thema vormden een goede basis voor het promotieonderzoek over kwaliteitsindicatoren voor de zorg voor mensen met downsyndroom, waarmee zij in 2015 startte en dat resulteerde in dit proefschrift. Het Jeroen Bosch Ziekenhuis in Den Bosch financierde dit promotieonderzoek, dat zij uitvoerde bij Tranzo, Wetenschappelijk centrum voor zorg en welzijn, Tilburg School of Social and Behavioral Sciences, Tilburg Universiteit. Sinds mei 2020 werkt Francine bij SKILZ (Stichting KwaliteitsImpuls Langdurige Zorg), een organisatie die evidence- en practice-based richtlijnen ontwikkelt voor professionals in de langdurige zorg voor ouderen en mensen met een verstandelijke beperking. Francine houdt van natuur en buitensport, en is een gepassioneerd maker (zang) van en luisteraar naar (klassieke) muziek. Ze woont in Arnhem, samen met haar partner Pieter en haar zonen Hugo, Philip en Otto.

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