Received: 7 March 2019 | Revised: 7 November 2019 | Accepted: 26 November 2019

DOI: 10.1111/jar.12692

ORIGINAL ARTICLE

JARID Journal of Applied Research in Intellectual Disabilities bild WILEY

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

Francine A. van den Driessen Mareeuw[1,2] | Antonia M. W. Coppus[3,4] | Diana M. J. Delnoij[5,6] | Esther de Vries[1,2]

[1]Tranzo, Scientific Center for Care and Wellbeing, Faculty of Social and Behavioral Sciences, Tilburg University, Tilburg, The Netherlands

[2]Jeroen Bosch Hospital, ME's-Hertogenbosch, The Netherlands

[3]Department for Primary and Community Care, Radboud University Medical Center, Nijmegen, The Netherlands

[4]Dichterbij, Center for the Intellectually Disabled, Gennep, The Netherlands

[5]Erasmus School of Health Policy and Management, Erasmus University, Rotterdam, The Netherlands

[6]National Health Care Institute, Diemen, The Netherlands

Correspondence
Francine A. van den Driessen Mareeuw, Tranzo, Scientific Center for Care and Wellbeing, Faculty of Social and Behavioral Sciences, Tilburg University, PO Box 90153, 5000 LE Tilburg, The Netherlands.
Email: f.a.vdndriessenmareeuw@uvt.nl

Funding information
Fonds NutsOhra, Grant/Award Number: 1403-029; Jeroen Bosch Hospital

Abstract

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

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

Results: According to the participants, healthcare 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 PDS.

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

KEYWORDS

Down syndrome, Netherlands, qualitative methods, quality of health care, quality of life

1 | INTRODUCTION

Down syndrome (DS) is associated with a large variety of health problems with varied severity and consequently complex healthcare needs, generally involving many different healthcare providers (Coppus, 2017; Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 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, Davidson, & Weintraub, 2013; Tenenbaum, Kastiel, Meiner, & Kerem, 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 intellectual disability physician (a medical doctor specialised in intellectual disability (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, Hollegien, Coppus, Delnoij, & de Vries, 2017), let alone how it is appraised by people with DS (PDS) and their caregivers (Barelds, van de Goor, van Heck, & Schols, 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 intellectual disability and DS (Bakker-van Gijssel et al., 2017; Graves et al., 2016; Kyrkou, 2018; van Schrojenstein Lantman-de Valk, Linehan, Kerr, & Noonan-Walsh, 2007), healthcare quality related to PDS has not been adequately researched (van den Driessen Mareeuw et al., 2017). Studies that do address quality in health care for PDS are traditionally conducted from a medical professional's perspective (Jensen & Davis, 2013; Jespersen, Michelsen, Holstein, Tjørnhøj-Thomsen, & Due, 2018; Phelps, Pinter, Lollar, Medlen, & Bethell, 2012). However, it is acknowledged increasingly that insight into the patient's perspective is crucial for improving healthcare quality (Poitras, Maltais, Bestard-Denommé, Stewart, & Fortin, 2018; Rathert, Wyrwich, & Boren, 2013), answering patients' needs (Barelds et al., 2010; Phelps et al., 2012; Trebble, Hansi, Hydes, Smith, & Baker, 2010), and increasing cost-effectiveness (Porter, 2010). Our aim is therefore to provide insight into the perspectives of PDS, parents and support staff regarding quality of health care for PDS in the Netherlands. This includes all primary and secondary health care that PDS may need during their lives (e.g., health care provided by paediatricians, intellectual disability physicians, physiotherapists and dieticians (within or outside Downteams), GPs). The present authors included PDS, their parents and support staff (i.e. people working in assisted living facilities for people with intellectual disability 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 healthcare 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 PDS, this system may involve parents and support staff, all playing a significant role in the lives of people with intellectual disability including DS (Mastebroek, Naaldenberg, van den Driessen Mareeuw, Lagro-Janssen, & van Schrojenstein Lantman-de Valk, 2016). Second, parents and support staff may complement PDS' views on healthcare quality or may function as proxies for PDS who are not able to verbally express themselves.

The World Health Organization (2006) identifies six dimensions of quality of care, being (a) effective (evidence-based and based on needs), (b) efficient (maximising resources, avoiding waste), (c) accessible (timely, geographically reasonable, in a suitable setting), (d) acceptable/patient-centred (taking into account preferences, culture of patient), (e) equitable (same level of quality for everyone) and (f) safe (minimising risk and harm). The present authors use these dimensions to study quality of health care for PDS. However, the present authors 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): (a) respect for patient's values, preferences and expressed needs, (b) information-education, (c) coordination and integration, (d) physical comfort, (e) emotional support and alleviation of fear/anxiety, (f) involvement of family/friends, (g) continuity and transition and (h) access (Rawson & Moretz, 2016; Singer et al., 2011).

Health (status) and (health-related) quality of life are considered to be important outcomes for assessing healthcare quality (Donabedian, 2005; Jespersen et al., 2018; Porter, 2010). Therefore, (health-related) quality of life is an important concept in the current study. The present authors 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 intellectual disability and include the following: (a) emotional well-being, (b) interpersonal relations, (c) material well-being, (d) personal development, (e) physical well-being, (f) self-determination, (g) social inclusion and (h) 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 PDS?*

- What are their experiences with received health care?
- How may health care influence the PDS' lives?"

2 | 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, Sainsbury, & Craig, 2007).

2.1 | 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). The present authors conducted semi-structured interviews with PDS and with parents of PDS, 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 21 August 2016 (no. EC-2016.21).

The research team consisted of a paediatrician with expertise in integrated care for PDS (professor) and data-driven research (EV), an expert in health services research (professor) and quality measurement (DD), an intellectual disability 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 intellectual disability (FDM).

2.2 | Participants

Purposive sampling was used to collect as many experiences, opinions and ideas about quality of health care for PDS 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; The present authors therefore included people ≥12 years with mild-to-moderate intellectual disability. The present authors 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 PDS with a larger age range (also younger than 12), and of PDS 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 intellectual disability, 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." The present authors 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 intellectual disability 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 PDS, 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 PDS, and/or arranged interviews with PDS, 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).

A total of 18 PDS 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 PDS and their parent(s) participated. In six cases, both PDS and their support staff participated. Characteristics of participants are shown in Table 1.

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

2.3 | 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" and 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 (e.g., 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 PDS lasted 30–75 min.

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 30–105 min.

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, and the other two focus groups had

TABLE 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 (years) mean [range]	31.7 [13–54]	57.3 [37–79]	39.8 [21–59]
Gender female; male	10; 8	14; 6 (five parent couples, nine mothers, one 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]) 24-hr support	3		
Level of intellectual disability[c]		n/a	n/a
Borderline (IQ70–85)	2		
Mild (IQ50–70)	8		
Moderate (IQ35–49)	7		
Severe (IQ20–34)[d]	1		
Health problems[c]			
Mentioned in number (and percentage) of interviews[e]		n/a	n/a
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%)		
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]) 24-hr support		1	9
Level of intellectual disability of child/client(s) with DS[c]	n/a		
Borderline (IQ70–85)		3	
Mild (IQ50–70)		4	8
Moderate (IQ35–49)		6	14
Severe (IQ20–34)		1	1
Not yet assessed (too young)		1	
Dementia			2
Health problems of child/client(s) with DS[c]			
Mentioned in number (and percentage) of total number of interviews or focus groups[e]	n/a		
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%)

(Continues)

TABLE 1 (Continued)

	Persons with DS (*n* = 18)	Parents/parent couples (*n* = 15)	Support staff (*n* = 35, supervising a total of 25 persons with DS)
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			
Female; male	n/a	7; 8	13; 12
Professional experience with PDS (years)			
<5	n/a	n/a	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 or focus groups.

participants from two organisations. Travelling costs to the venue where the focus groups took place were reimbursed. The focus groups took about 30 min to 2 hrs (depending on time available by participating support staff), and the single interview lasted 50 min.

The interviews with PDS 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.

2.4 | Topics discussed

An interview or focus group guide was composed for each specific group of participants (PDS, parents and support staff). The different guides included similar topics based upon the eight domains of quality of life as formulated by Schalock et al. (2005) and patients' experiences (in this case of PDS, 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 PDS included pictures (of e.g., healthcare providers) and pictograms (e.g., representing abstract concepts like "sad" or "bored"). A draft of the interview guide for PDS was discussed (and adapted accordingly) with other researchers with experience in interviewing people with mild-to-moderate intellectual disability. A summary of the interview guides and some example questions are presented in Appendix 1.

2.5 | 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, Heath, Cameron, Rashid, & Redwood, 2013), see Table 2. All authors were involved in data analysis (including coding). To

TABLE 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 pre-defined codes) coding (Gale et al., 2013), which ensured that important themes in the data were not missed and enabled structuring the complex data. Pre-defined codes derived from theory: quality of life domains (Schalock et al., 2005), quality of care dimensions (WHO, 2006) and 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 2) 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), allowing interpretation

maximise objective analysis, one-third of the transcripts were double-coded by two authors (by FDM and AC, DD, and EV, respectively). Data were managed using the software package Atlas.ti 8 for Windows.

3 | RESULTS

In describing the results, the present authors use "participants with DS/ PDS" or "parents" if the present authors mean (parents of) PDS 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.

3.1 | 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 PDS (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 (55 yrs) of woman with DS (23 yrs)). Parents either indicated that health problems were managed well, generally resulting in a low burden, or experienced difficulties with managing the complex healthcare needs. Support staff too considered PDS 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, 2017).

3.2 | Healthcare utilisation and "Downteams"

According to participants with DS, parents and support staff, PDS received, or had received, care by a large variety of healthcare providers. Roughly spoken, the paediatrician and speech therapist were visited during childhood; intellectual disability 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 PDS who are less able to display pain or other symptoms of disease. The reasons mentioned by parents are in accordance with 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 healthcare 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 PDS, 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 intellectual disability instead of, e.g., in a general hospital and/or within the community.

3.3 | Role of parents and support staff

Participants with DS, parents and support staff reported that PDS generally needed support deciding about visiting a doctor, making appointments with healthcare professionals, communicating during consultations, and sharing health or treatment information with (other) healthcare professionals, support staff, parents or other relatives. This is in line with literature on adults with intellectual disability in primary care (Mastebroek et al., 2016). When PDS were living with their parents, parents offered this support. PDS 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 healthcare providers on their own. Parents and support staff stressed that especially in such cases, it is important that healthcare 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).

3.4 | Perceived healthcare quality

Generally, participants with DS, parents and support staff qualified health care for PDS as good, although less positive stories also were heard regarding health care for (especially) adult PDS, including rude healthcare 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 healthcare 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 healthcare quality, especially regarding adult PDS, 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 healthcare 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 adult PDS 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 (49 yrs) of a boy with DS (13 yrs)). Similar time and energy constraints are reflected in literature (Phelps et al., 2012; Povee, Roberts, Bourke, & Leonard, 2012).

3.5 | 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 PDS. Parents and support staff underlined that healthcare 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 intellectual disability in other studies (Kyrkou, 2014; Minnes & Steiner, 2009). According to parents and support staff, applying a holistic approach also means that healthcare 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 adult PDS: sleep apnoea (impaired daily functioning and behaviour, not always detected), communication problems (impedes emotional expression and social interaction) and 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 healthcare 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 (54 yrs) of a boy with DS (14 yrs)). Goodman and Brixner (2013) confirm the importance of considering the impact of a treatment on quality of life in PDS.

3.6 | Adapted communication, trust and respect

Specific communication difficulties, such as language processing or hearing problems, commonly present among PDS (Grieco et al., 2015) may hinder communication between healthcare professionals

and their patients with DS. Adult participants with DS argued that healthcare 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 yrs)). 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 PDS 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 (54 yrs) of a boy with DS (14 yrs)). Similar issues were found in studies on health care for people with intellectual disability (including DS) (Mastebroek et al., 2016; Miller et al., 2009). However, in PDS, these issues may need even more attention, because communication difficulties are prominent among PDS and they may have different cognitive and behavioural profiles, including different pain representation, compared to people with intellectual disability (Grieco et al., 2015; Kyrkou, 2014).

3.7 | Complexity of (health)care

Although participants considered healthcare 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 [PDS] 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 (49 yrs) of a boy with DS (13 yrs)). 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 (57 yrs) about her son (man with DS (26 yrs)) 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 (63 yrs) of a woman with DS (28 yrs)). Minnes and Steiner (2009) also observed this "stress in dealing with the healthcare system and in negotiating relationships with practitioners."

There were also parents of PDS 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 (37 yrs) of a girl with DS (7 yrs)). 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.

3.8 | A need for coordination

According to parents and support staff, collaboration and good communication between all the different professionals involved are important elements of healthcare quality. This notion is supported by literature on the topic (Kyrkou, 2014; Miller et al., 2009). 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 healthcare 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 (57 yrs) of a man with DS (25 yrs)). According to parents, this professional should also connect with actors outside health care, for example school, daily activity centre and social services. This coordinating role was not allocated to a specific professional, but could be, or was, fulfilled by a paediatrician, GP, intellectual disability 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 (57 yrs) of a man with DS (25 yrs)). The importance of smooth transitions, good coordination and continuity is confirmed in literature (Dyke, Bourke, Llewellyn, & Leonard, 2013; Kyrkou, 2014; Miller et al., 2009; Woodward, Swigonski, & Ciccarelli, 2012).

4 | DISCUSSION

The present authors explored what PDS and their representatives (parents and support staff) consider to be healthcare quality and how this may impact PDS' quality of life. In summary, PDS stressed the importance of healthcare 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 healthcare quality. Support staff complemented that for PDS 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 healthcare quality in general (not DS-specific) (Di Blasi, Harkness, Ernst, Georgiou, & Kleijnen, 2001; Morgan & Yoder, 2012). However, it is argued that compared to the general population, and to people with intellectual disability, PDS 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 healthcare requirements. First, according to participating parents, benefits and burden of a treatment may be different for PDS compared to the general population. This means that healthcare professionals should determine the best outcome (low burden, high benefit), by considering DS-specific conditions, and acknowledging the living/family situation of PDS and stress experienced by families. Second, the specific profile of PDS requires adapted professional–patient interaction. Therefore, healthcare 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 healthcare 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, Gorawara-Bhat, & Lamb, 2000; Stewart et al., 2000). Third, the care and support system is complex and includes a specific combination of a large number of healthcare and other professionals. Coordinating this complex system around children and adults with DS requires good management skills of parents/ other carers of PDS. 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 healthcare 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 PDS, 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, Schepman, Opheij, & Bruijnzeels, 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, Calciolari, Goodwin, & Stein, 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, Luijkx, Elissen, & Vrijhoef, 2016; Van Duijn, Zonneveld, Montero, Minkman, & Nies, 2018; González-Ortiz 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 PDS. 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).

4.1 | Strengths and limitations

This study's strength is that it investigated healthcare quality through the eyes of PDS and their caregivers. This perspective is

crucial in determining what person-centred care for PDS really should be, which is a requirement for improving healthcare quality. Another strength is that the present authors included (parents and support staff of) children and adults with DS. The findings are therefore sensitive to healthcare 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, PDS 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 PDS 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.

5 | CONCLUSION

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

ACKNOWLEDGMENTS

The authors would like to thank all participants in the study for their time and willingness to share their opinions and experiences. Additionally, the authors acknowledge the help from the Dutch Down Syndrome Association ("Stichting Downsyndroom"), service providers and others in recruiting participants. The authors would also like to thank Rosemary van der Avort-Lier for her assistance by the editing of this paper as native English speaker. This work was financially supported by the Fonds NutsOhra (fund), Amsterdam, the Netherlands [grant number: 1403-029]; and the Jeroen Bosch Hospital, 's-Hertogenbosch, the Netherlands.

CONFLICTS OF INTEREST

The authors declare no potential conflicts of interest.

ORCID

Francine A. van den Driessen Mareeuw https://orcid.org/0000-0002-4837-5261

REFERENCES

Bakker-van Gijssel, E. J., Olde Hartman, T. C., Lucassen, P. L. B. J., van den Driessen Mareeuw, F., Dees, M. K., Assendelft, W. J. J., & van Schrojenstein Lantman-de Valk, H. M. J. (2017). GPs' opinions of health assessment instruments for people with intellectual disabilities: A qualitative study. *British Journal of General Practice*, *67*, e41–e48. https://doi.org/10.3399/bjgp16X688585

Barelds, A., van de Goor, I., van Heck, G., & Schols, J. (2010). Quality of care and service trajectories for people with intellectual disabilities: Defining the aspects of quality from the client's perspective. *Scandinavian Journal of Caring Sciences*, *24*(1), 164–174. https://doi.org/10.1111/j.1471-6712.2009.00701.x

Busetto, L., Luijkx, K. G., Elissen, A. M. J., & Vrijhoef, H. J. M. (2016). Context, mechanisms and outcomes of integrated care for diabetes mellitus type 2: A systematic review. *BMC Health Services Research*, *16*(18), 1–14. https://doi.org/10.1186/s12913-015-1231-3

Coppus, A. (2017). Comparing generational differences in persons with Down syndrome. *Journal of Policy and Practice in Intellectual Disabilities*, *14*(2), 118–123. https://doi.org/10.1111/jppi.12214

Di Blasi, Z., Harkness, E., Ernst, E., Georgiou, A., & Kleijnen, J. (2001). Influence of context effects on health outcomes: A systematic review. *The Lancet*, *357*(9258), 757–762. https://doi.org/10.1016/S0140-6736(00)04169-6

Dimitropoulos, G., Morgan-Maver, E., Allemang, B., Schraeder, K., Scott, S. D., Pinzon, J., Samuel, S. (2019). Health care stakeholder perspectives regarding the role of a patient navigator during transition to adult care. *BMC Health Services Research*, *19*(390), 1–10. https://doi.org/10.1186/s12913-019-4227-6

Donabedian, A. (2005). Evaluating the quality of medical care. *The Millbank Quarterly*, *83*(4), 691–729. https://doi.org/10.1111/.1468-0009.2005.00397.x

Dyke, P., Bourke, J., Llewellyn, G., & Leonard, H. (2013). The experiences of mothers of young adults with an intellectual disability transitioning from secondary school to adult life. *Journal of Intellectual and Developmental Disability*, *38*(2), 149–162. https://doi.org/10.3109/13668250.2013.789099

Gale, N. K., Heath, G., Cameron, E., Rashid, S., & Redwood, S. (2013). Using the framework method for the analysis of qualitative data in multi-disciplinary health research. *BMC Medical Research Methodology*, *13*(1), 117. https://doi.org/10.1186/1471-2288-13-117

González-Ortiz, L. G., Calciolari, S., Goodwin, N., & Stein, V. (2018). The core dimensions of integrated care: A literature review to support the development of a comprehensive framework for implementing integrated care. *International Journal of Integrated Care*, *18*(3), 1–12. https://doi.org/10.5334/ijic.4198

Goodman, M. J., & Brixner, D. I. (2013). New therapies for treating Down syndrome require quality of life measurement. *American Journal of Medical Genetics Part A*, *161*(4), 639–641. https://doi.org/10.1002/ajmg.a.35705

Graves, R. J., Graff, J. C., Esbensen, A. J., Hathaway, D. K., Wan, J. Y., & Wicks, M. N. (2016). Measuring health-related quality of life

of adults with Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, *121*(4), 312–326. https://doi.org/10.1352/1944-7558-121.4.312

Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). *Down syndrome: Cognitive and behavioral functioning across the lifespan*. Paper presented at the American Journal of Medical Genetics Part C: Seminars in Medical Genetics. https://doi.org/10.1002/ajmg.c.31439

Jensen, K. M., & Davis, M. M. (2013). Health care in adults with Down syndrome: A longitudinal cohort study. *Journal of Intellectual Disability Research*, *57*(10), 947–958. https://doi.org/10.1111/j.1365-2788.2012.01589.x

Jespersen, L. N., Michelsen, S. I., Holstein, B. E., Tjørnhøj-Thomsen, T., & Due, P. (2018). Conceptualization, operationalization, and content validity of the EQOL-questionnaire measuring quality of life and participation for persons with disabilities. *Health and Quality of Life Outcomes*, *16*(1), 199. https://doi.org/10.1186/s12955-018-1024-6

Kinnear, D., Morrison, J., Allan, L., Henderson, A., Smiley, E., & Cooper, S.-A. (2018). Prevalence of physical conditions and multimorbidity in a cohort of adults with intellectual disabilities with and without Down syndrome: Cross-sectional study. *British Medical Journal Open*, *8*(2), e018292.

Kyrkou, M. R. (2014). Health-related quality of life. *International Public Health Journal*, *6*(4), 355–370.

Kyrkou, M. R. (2018). Health-related family quality of life when a child or young person has a disability. *International Journal of Child, Youth and Family Studies*, *9*(4), 49–74 http://dx.doi.org/10.18357/ijcyfs94201818640

Levinson, W., Gorawara-Bhat, R., & Lamb, J. (2000). A study of patient clues and physician responses in primary care and surgical settings. *JAMA*, *284*(8), 1021–1027. https://doi.org/10.1001/jama.284.8.1021

Mastebroek, M., Naaldenberg, J., van den Driessen Mareeuw, F. A., Lagro-Janssen, A. L., & van Schrojenstein Lantman-de Valk, H. M. (2016). Experiences of patients with intellectual disabilities and carers in GP health information exchanges: A qualitative study. *Family Practice*, *33*(5), 543–550. https://doi.org/10.1093/fampra/cmw057

Miller, A. R., Condin, C. J., McKellin, W. H., Shaw, N., Klassen, A. F., & Sheps, S. (2009). Continuity of care for children with complex chronic health conditions: Parents' perspectives. *BMC Health Services Research*, *9*(1), 242. https://doi.org/10.1186/1472-6963-9-242

Minnes, P., & Steiner, K. (2009). Parent views on enhancing the quality of health care for their children with fragile X syndrome, autism or Down syndrome. *Child: Care, Health and Development*, *35*(2), 250–256. https://doi.org/10.1111/j.1365-2214.2008.00931.x

Morgan, S., & Yoder, L. H. (2012). A concept analysis of person-centered care. *Journal of Holistic Nursing*, *30*(1), 6–15. https://doi.org/10.1177/0898010111412189

Phelps, R. A., Pinter, J. D., Lollar, D. J., Medlen, J. G., & Bethell, C. D. (2012). Health care needs of children with Down syndrome and impact of health system performance on children and their families. *Journal of Developmental and Behavioral Pediatrics*, *33*(3), 214–220. https://doi.org/10.1097/DBP.0b013e3182452dd8

Poitras, M.-E., Maltais, M.-E., Bestard-Denommé, L., Stewart, M., & Fortin, M. (2018). What are the effective elements in patient-centered and multimorbidity care? A scoping review. *BMC Health Services Research*, *18*(1), 446. https://doi.org/10.1186/s12913-018-3213-8

Porter, M. E. (2010). What is value in health care? *New England Journal of Medicine*, *363*(26), 2477–2481. https://doi.org/10.1056/NEJMp1011024

Povee, K., Roberts, L., Bourke, J., & Leonard, H. (2012). Family functioning in families with a child with Down syndrome: A mixed methods approach. *Journal of Intellectual Disability Research*, *56*(10), 961–973. https://doi.org/10.1111/j.1365-2788.2012.01561.x

Rathert, C., Wyrwich, M. D., & Boren, S. A. (2013). Patient-centered care and outcomes: A systematic review of the literature. *Medical Care Research and Review*, *70*(4), 351–379. https://doi.org/10.1177/1077558712465774

Rawson, J. V., & Moretz, J. (2016). Patient-and family-centered care: A primer. *Journal of the American College of Radiology*, *13*(12), 1544–1549. https://doi.org/10.1016/j.jacr.2016.09.003

Schalock, R. L., Verdugo, M. A., Jenaro, C., Wang, M., Wehmeyer, M., Jiancheng, X., & Lachapelle, Y. (2005). Cross-cultural study of quality of life indicators. *American Journal on Mental Retardation*, *110*(4), 298–311. https://doi.org/10.1352/0895-8017(2005)110[298:CSOQOL]2.0.CO;2

Simões, C., & Santos, S. (2016). Comparing the quality of life of adults with and without intellectual disability. *Journal of Intellectual Disability Research*, *60*(p4), 378–388. https://doi.org/10.1111/jir.12256

Singer, S. J., Burgers, J., Friedberg, M., Rosenthal, M. B., Leape, L., & Schneider, E. (2011). Defining and measuring integrated patient care: Promoting the next frontier in health care delivery. *Medical Care Research and Review*, *68*(1), 112–127. https://doi.org/10.1177/1077558710371485

Skotko, B. G., Davidson, E. J., & Weintraub, G. S. (2013). Contributions of a specialty clinic for children and adolescents with Down syndrome. *American Journal of Medical Genetics Part A*, *161*(3), 430–437. https://doi.org/10.1002/ajmg.a.35795

Stewart, M., Brown, J. B., Donner, A., McWhinney, I. R., Oates, J., Weston, W. W., & Jordan, J. (2000). The impact of patient-centered care on outcomes. *Journal of Family Practice*, *49*(9), 796–796.

Tavakol, M., & Sandars, J. (2014). Quantitative and qualitative methods in medical education research: AMEE Guide No 90: Part II. *Medical Teacher*, *36*(10), 838–848. https://doi.org/10.3109/0142159X.2014.915297

Tenenbaum, A., Kastiel, Y., Meiner, Z., & Kerem, E. (2008). Multidisciplinary care of persons with Down syndrome in Jerusalem. *International Journal on Disability and Human Development*, *7*(3), 355–358. https://doi.org/10.1515/IJDHD.2008.7.3.355

Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): A 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*, *19*(6), 349–357. https://doi.org/10.1093/intqhc/mzm042

Trebble, T. M., Hansi, N., Hydes, T., Smith, M. A., & Baker, M. (2010). Process mapping the patient journey through health care: An introduction. *BMJ*, *341*(7769), 394–397. https://doi.org/10.1136/bmj.c4078

UN. (2006). *Convention on the rights of persons with disabilities*. New York, NY: UN.

Valentijn, P. P., Schepman, S. M., Opheij, W., & Bruijnzeels, M. A. (2013). Understanding integrated care: A comprehensive conceptual framework based on the integrative functions of primary care. *International Journal of Integrated Care*, *13*, https://doi.org/10.5334/ijic.886

van den Driessen Mareeuw, F. A., Hollegien, M. I., Coppus, A. M., Delnoij, D. M., & de Vries, E. (2017). In search of quality indicators for Down syndrome healthcare: A scoping review. *BMC Health Services Research*, *17*(1), 284. https://doi.org/10.1186/s12913-017-2228-x

van Duijn, S., Zonneveld, N., Montero, A. L., Minkman, M., & Nies, H. (2018). Service integration across sectors in Europe: Literature and practice. *International Journal of Integrated Care*, *18*(2), 1–13. https://doi.org/10.5334/ijic.3107

van Schrojenstein Lantman-de Valk, H., Linehan, C., Kerr, M., & Noonan-Walsh, P. (2007). Developing health indicators for people with intellectual disabilities. The method of the Pomona project. *Journal of Intellectual Disability Research*, *51*(6), 427–434. https://doi.org/10.1111/j.1365-2788.2006.00890.x

Weijerman, M. E., & De Winter, J. P. (2010). Clinical practice. *European Journal of Pediatrics*, *169*(12), 1445–1452. https://doi.org/10.1007/s00431-010-1253-0

WHO. (2006). *Quality of care: A process for making strategic choices in health systems*. Geneva, Switzerland: Wold Health Organization.

WHO Europe. (2016). *Integrated care models: An overview*. Copenhagen, Denmark: World Health Organization Regional Office for Europe.

Wiering, B. M., Noordman, J., Tates, K., Zwaanswijk, M., Elwyn, G., De Bont, E. S. J. M., … Van Dulmen, S. (2016). Sharing decisions during diagnostic consultations; an observational study in pediatric oncology. *Patient Education and Counseling*, *99*(1), 61–67. https://doi.org/10.1016/j.pec.2015.07.026

Woodward, J. F., Swigonski, N. L., & Ciccarelli, M. R. (2012). Assessing the health, functional characteristics, and health needs of youth attending a noncategorical transition support program. *Journal of Adolescent Health*, *51*(3), 272–278. https://doi.org/10.1016/j.jadohealth.2011.12.016

How to cite this article: van den Driessen Mareeuw FA, Coppus AMW, Delnoij DMJ, de Vries E. Quality of health care according to people with Down syndrome, their parents and support staff—A qualitative exploration. *J Appl Res Intellect Disabil*. 2019;00:1–19. https://doi.org/10.1111/jar.12692

APPENDIX 1

Overview of interview/focus group guides and example questions

	Examples of questions in		
Topic	**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 healthcare providers did your son/daughter visit in his/her life?	Please mention one healthcare 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)
Healthcare 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 healthcare for people with DS?	What is the first thing that comes in mind when you think about quality in healthcare 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 2

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)
Healthcare 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

Framework matrix

	Said by:			
Theme	People with DS	Parents	Support staff	Interpretation by authors
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 do not 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)

(Continues)

APPENDIX 3 (Continued)

Theme	Said by: People with DS	Parents	Support staff	Interpretation by authors
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 are not). Parents and support staff try to create "normal lives."
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 is 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

(Continues)

APPENDIX 3 (Continued)

Theme	Said by: People with DS	Parents	Support staff	Interpretation by authors
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 do not 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
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." Healthcare 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 is 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. • Healthcare professionals should acknowledge the challenges of having a child with DS

(Continues)

APPENDIX 3 (Continued)

Theme	Said by: People with DS	Parents	Support staff	Interpretation by authors
Patient journey	• Various aids are being used, often mentioned: glasses, arch support. Not always accepted. • Various medications • Various healthcare 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
Quality of care: Person-centred care: general	"If you know a healthcare professional for some time, you can trust that person, which makes talking easier"	• Trust relationship with healthcare professional is important. • Waiting (waiting room) may be difficult for people with DS (e.g., because they become nervous). • Healthcare professionals have to "click" with a person with DS. • Healthcare professional who knows about all health problems and can give advice about all conditions together is needed	A known healthcare professional is important	• Trust relationship and knowing each other is important: need for continuity of care. • "Clicking" of healthcare professionals and person with DS is important. • Healthcare 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	Healthcare professional has to put me at ease, make jokes and ask "other" (non-medical) questions	Healthcare 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 healthcare professional talks slowly, I can understand him/her better	• Healthcare 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	• Healthcare 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. • Healthcare 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		• Healthcare professional taking role of "sparring partner" is nice. • Healthcare 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, healthcare professionals and support staff is needed
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

(Continues)

APPENDIX 3 (Continued)

Theme	Said by: People with DS	Parents	Support staff	Interpretation by authors
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. • Healthcare professional should consider pressure experienced by parents related to managing care. • Parents prefer a holistic approach	• Healthcare 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." • Healthcare professionals should try to align their actions with internal world of people with DS
Quality of care: Effectiveness	The doctor has to cure me	• Healthcare professional does not always have DS-specific knowledge, but does not always need to have it. As long as (s)he really "sees" the person with DS. • Many healthcare 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, and we will go to a doctor if needed. • Early intervention ® often used for development of children with DS. • Support staff does not 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 • Healthcare 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
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 healthcare 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 healthcare providers refuse to treat people with DS. • Special treatment is not always needed (people with DS are also just people)	• Some healthcare professionals do not 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

(Continues)

APPENDIX 3 (Continued)

Theme	Said by: People with DS	Parents	Support staff	Interpretation by authors
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
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 is 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")
Information lack, finding and exchange.		• Would have liked more information on impact of DS on family, educational support and school choice. • It is often difficult to find right healthcare 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 healthcare 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

(Continues)

APPENDIX 3 (Continued)

Theme	Said by: People with DS	Parents	Support staff	Interpretation by authors
How quality of care may influence quality of life	• Sleep apnoea has been treated, less tired now • Some think it is 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

Note: 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).