Towards seamless and sustainable care for Parkinson's disease

FLORIS P. VLAANDEREN

TOWARDS SEAMLESS AND SUSTAINABLE CARE FOR PARKINSON'S DISEASE

Proefschrift

ter verkrijging van de graad van doctor
aan de Radboud Universiteit Nijmegen
op gezag van de rector magnificus prof. dr. J.H.J.M. van Krieken,
volgens besluit van het college van decanen
in het openbaar te verdedigen op woensdag 27 oktober 2021
om 14.30 uur precies

door

Floris Pieter Vlaanderen
geboren op 23 oktober 1987
te Amsterdam

Towards seamless and sustainable care for Parkinson's disease

Design/Lay-out
Proefschriftenbalie, Nijmegen

Print
Ipskamp Printing, Enschede

ISBN 978-94-6421-502-1

Promotoren
Prof. dr. B.R. Bloem
Prof. dr. P.P.T. Jeurissen

Copromotoren
Dr. M.J. Meinders
Dr. M.A.C. Tanke

Manuscriptcommissie
Prof. dr. W.J.J. Assendelft
Prof. dr. M.M. Rovers
Dr. J.P.P. van Vugt (Medisch Spectrum Twente)

CONTENTS

CHAPTER 1

General introduction

1.1 PARKINSON'S DISEASE

Parkinson's disease (PD) is a chronic progressive neurodegenerative disorder, characterized by a wide range of motor and nonmotor symptoms (Lees, Hardy, and Revesz 2009) (see Box 1.1). Despite the invariably progressive character of PD, the life expectancy of people with PD is only modestly influenced by the disease (de Lau et al. 2005). Most people with PD live for many years with their disease; although with progressive reductions in quality of life. PD is relatively common: the reported prevalence in the general population is around 0.3%, but this rises rapidly with age – up to 1.4% for over 55s (Kowal et al. 2013), with an average age at diagnosis of 70.5 years (Van Den Eeden et al. 2003). The prevalence of PD in 2040 is estimated to be be twice as high as in 2015 (Dorsey et al. 2007; Dorsey et al. 2018; Dorsey et al. 2020); in part as a result of the aging of the population, but also because of exposure to abundantly present environmental toxins, such as pesticides or trichloroethylene (Dorsey et al. 2007; Dorsey et al. 2018; Dorsey et al. 2020). Both the incidence and prevalence seem to be higher in men than in women (Georgiev et al. 2017).

Box 1.1: Symptoms of Parkinson's disease

Besides the four traditional motor symptoms of Parkinson's disease (tremor, bradykinesia, rigidity and postural instability), many other motor and non-motor symptoms may occur, resulting in a complex phenotype that requires a comprehensive multidisciplinary management (see Table 1.1) (Chou, Hurtig, and Eichler 2019).

TABLE 1.1 | **Symptoms of PD**

Motor symptoms	Non-motor symptoms
- Tremor	- Cognitive dysfunction
- Bradykinesia	- Psychosis
- Rigidity	- Mood disorders (depression, anxiety, apathy/abulia)
- Postural instability	- Sleep disturbance
- Masked facial expression	- Fatigue
- Speech disturbances	- Autonomic dysfunction
- Dysphagia and sialorrhea	- Olfactory dysfunction
- Impaired vision	- Pain and sensory disturbances
- Micrographia	- Dermatologic findings (seborrhea)
- Dystonia	
- Stooped posture	
- Shuffling, short-stepped gait	
- Freezing and festination	

There is currently no cure for PD; nor can the disease progression be slowed down. The treatment of PD is therefore entirely symptomatic, and aims to optimize the person's quality of life by reducing the symptom burden, and avoiding PD-related complications such as falls and fractures. Treatment includes pharmacotherapy, various allied healthcare treatments and several device-aided treatments, including deep brain stimulation and two types of pump therapies (Hijdra, Koudstaal, and Roos 2016). The costs of these treatments are substantial: in the Netherlands, the healthcare costs of the approximately 40,000 people with PD were estimated to be at least € 204.1 million in 2017 (National Institute of Public Health and the Environment 2019a, 2019c). This figure does not include the costs of PD-related complications or comorbidities; neither does it include the secondary economic costs of the disease (for example: loss of productivity of the people with PD or their family members)(National Institute of Public Health and the Environment 2019b). Therefore, the total costs of PD are undoubtedly even considerably higher.

1.2 SEAMLESS AND SUSTAINABLE CARE FOR PEOPLE WITH PD

To achieve the best results from the previously mentioned treatments, the care for people with PD needs to be well organised and coordinated. This is often referred to as 'seamless' care (Hammond 2010). Seamless care is care which is consistent and coherent, marked by an orderly, logically and aesthetically consistent relation of parts, without discontinuities or disparities, uniform in quality and combined in an inconspicuous way (Hammond 2010) (Box 1.2). Key objectives are continuity of care, multiple uses of healthcare data, and collaborative partnerships between healthcare providers (Hammond 2010; Howitt 2011). The concept of seamless care is applied when there is a transition of a patient from one healthcare provider to another, most notably from the hospital to the persons' own home (Spehar et al. 2005; Knight et al. 2013). Such transitions are very common in the overall management process for people living with PD, with involvement of multiple healthcare providers who work in different echelons of healthcare. Even people with relatively mild PD regularly visit general practitioners, neurologists, specialized nurses, physiotherapists, occupational therapists and speech-language therapists. In the more advanced stage, dieticians, psychologists, other medical specialists and nursing home facilities often also become involved (Bloem et al. 2010a). The number of different healthcare providers increases even more when complications occur, such as a hip fracture. Regrettably, these many providers rarely work together as a team; and indeed, many providers do not well know what others can offer to the overall management process. Sharing of information is generally poor, which hampers effective care delivery. These considerations highlight the importance of ascertaining a more seamless care delivery for people living with PD.

Apart from being seamless, the care for people with PD also needs to be sustainable. Sustainable healthcare implies that the level of care can be maintained through time, and that the care is efficient and does not unnecessary drain finite resources (Jeurissen, Maarse, and Tanke 2018) (Box 1.2). Since life expectancy of the general population is rising, people use more care and for a longer period. This causes pressure on healthcare costs, which are rising globally (Xu et al. 2018). PD is no exception, since the incidence of PD increases dramatically over the age of 60 (Tysnes and Storstein 2017).

Box 1.2: Definitions of seamless and sustainable care

Seamless care: care which is consistent and coherent, marked by an orderly, logically and aesthetically consistent relation of parts, without discontinuities or disparities, uniform in quality and combined in an inconspicuous way (Hammond 2010).

Sustainable care: care which can be maintained through time, and that is efficient and does not unnecessary drain ending resources (Jeurissen, Maarse, and Tanke 2018).

1.3 MANY PEOPLE WITH PD DO NOT RECEIVE OPTIMAL CARE

Regrettably, the current care provision for people with PD is often far from seamless and sustainable. First, the care people with PD receive varies much between persons, thus not 'consistent', 'coherent' or 'uniform in quality'. For example, approximately only half of the Europeans with PD regularly visit a neurologist (Bloem and Stocchi 2015). In the USA, forty-two percent of the people with PD over 65 years of age do not see a neurologist at all, and this percentage can be as high as 100% in some rural areas (Willis et al. 2011). Such variation in accessibility of neurologists also affects the utilization of other healthcare services. For example, neurologist visits seem to be an important driver for rehabilitation therapy use among elderly with PD in the USA (Fullard et al. 2017). Referrals to allied healthcare providers also vary between different countries (Stocchi and Bloem 2013). Besides, allied healthcare services are inappropriately allocated: a survey among Dutch people with PD in 2004 revealed that 41% of those requiring a referral to a physiotherapist did not receive physiotherapy. In contrast, half of the people with PD who did not require physiotherapy received it anyway (Keus et al. 2004). Suboptimal allocation of physiotherapy resources can lead to more complications, such as hip fractures, among people with PD (Bloem et al. 2017). Potentially avoidable complications not only affect the involved people with PD, but ultimately also society, as these often require costly treatments, affecting the sustainability of care.

Second, many studies show that the current care for people with PD is not 'marked by an orderly, logically and aesthetically consistent relation of parts'. In a European survey, forty-five percent of respondents with PD regarded the diagnostic process as 'poor' or 'very poor' (Bloem and Stocchi 2012). Dutch people with PD reported lack of emotional support from their healthcare providers (van der Eijk, Faber, Al Shamma, et al. 2011). Additionally, people with PD often feel not involved in treatment decisions. Although in the above-mentioned European survey 63% of respondents with PD were satisfied with the attention from healthcare providers, just 12% felt involved in treatment decisions (Bloem and Stocchi 2015). While many people with PD desire more active involvement in treatment decisions, they lack sufficient tools and information to do so (van der Eijk, Faber, Al Shamma, et al. 2011). Thus, they receive care focussed on pharmacological suppression of symptoms, and not on what really matters for the quality of life of the individual person with PD (van der Eijk, Faber, Al Shamma, et al. 2011; Findley and Baker 2002; Grosset and Grosset 2005).

Third, financial issues in current healthcare systems might stand in the way of seamless and sustainable care delivery. Separate payment systems between different echelons of the healthcare sector – e.g., between hospital care, long-term care or primary care – frustrate continuity of care, hamper a shared use of healthcare data, and interfere with an effective collaboration between healthcare providers (Jeurissen, Maarse, and Tanke 2018). In addition, the dominant fee-for-service system harms sustainability by inducing healthcare providers to increase their volumes; i.e., the number of treatments they can claim. More care delivery does not necessarily mean better care: incentivising volume can result in overuse of more or less expensive treatments that are not in line with what people with PD desire (Jeurissen, Maarse, and Tanke 2018). Fee-for-service can also frustrate a more home-based care approach, since healthcare providers usually lose income when facilitating such care (Landers et al. 2016).

Finally, the caseload for PD is generally low per individual healthcare provider. The care for people with PD is typically delivered by scattered healthcare providers who each manage only very few people with PD, with possible adverse effects. Specifically, this lack of concentration of care might harm seamless care delivery in two ways. First, it might prevent adequate coordination and multidisciplinary collaboration, and lead to lack of alignment between different treatments (Nijkrake et al. 2009; van der Eijk, Faber, Al Shamma, et al. 2011). Second, the knowledge about and expertise in PD might fall short. Many people with PD indeed encounter difficulties in finding healthcare providers with an adequate level of expertise in PD (Keus et al. 2004; van der Eijk, Faber, Al Shamma, et al. 2011). Several surveys among allied healthcare providers confirm concerns about a lack of expertise among physiotherapists, occupational therapists and speech-language therapists (Nijkrake et al. 2009).

Studying these elements of PD care might result in suggestions for improvements, and thereby contribute to more seamless and sustainable care. We will analyse these elements from the perspective of the person with PD, as well as the perspective of society.

1.4 TOWARDS SEAMLESS AND SUSTAINABLE CARE FOR PEOPLE WITH PD

Initiatives have been taken to make the care for people with PD more seamless and sustainable. Some examples are the Centers of Excellence network of the Parkinson's Foundation (Parkinson's Foundation 2019a, 2019b), the World Parkinson's Program (Worlds Parkinson's Program 2019), and the UK Parkinson's Excellence Network (Parkinson's UK 2019). All these initiatives enhance seamless and sustainable care by researching and improving expertise and multi-disciplinary collaboration of healthcare providers involved in PD.

Since this thesis is conducted in the Dutch care setting, it is important to assess the Dutch situation. In the Netherlands, the initiative with the greatest impact on PD care is ParkinsonNet, a not-for-profit healthcare organisation exclusively for people with PD or a form of atypical parkinsonism (Bloem and Munneke 2014). Founded in 2004, the ParkinsonNet approach strives to improve the care for people with PD by improving multi-disciplinary collaboration, by enhancing PD-specific knowledge among both healthcare providers and people with PD, and by stimulating patient-centeredness. To achieve this, ParkinsonNet has created regional networks of a limited number of highly motivated and well-trained healthcare providers, who each treat large numbers of people with PD. These regional networks include neurologists, physiotherapists, occupational therapists, speech & language therapists, dieticians, specialised Parkinson nurses, elderly care specialists, rehabilitation specialists, psychiatrists, psychologists, pharmacists, social workers, and sex therapists (Bloem and Munneke 2014; Bloem et al. 2017). These providers receive special training based on national and international clinical guidelines; this includes both a three-day basic training course and annual follow-up training courses. Expertise is further enhanced by raising the caseload per provider. Additionally, different ICT innovations have been implemented to support both providers and people with PD. Examples of these are web-based communities, ParkinsonTV (a web-based educational television programme) and a web-based search engine to readily identify specialised ParkinsonNet providers (Bloem and Munneke 2014). An overview of the key components of ParkinsonNet is provided in Table 1.2.

Today, ParkinsonNet consists of 70 regional networks with over 3,200 specialised healthcare providers (ParkinsonNet 2019). The approach has been evaluated in

TABLE 1.2 | **Key components of the ParkinsonNet approach (Bloem and Munneke 2014)**

Guidelines:	Development of mono- and multidisciplinary guidelines
Selection:	Inclusion of a restricted number of participants
Preferred referral:	Preferred referral to ParkinsonNet participants to increase caseload
Education:	Baseline training for participants and knowledge exchange structures
Commitment:	Stimulating adherence to guidelines
Transparency:	Open source publication of outcomes
Patient centeredness:	Empowered by internet communities and questionnaires
ICT platform:	Innovative telehealth solutions

multiple studies (Nijkrake et al. 2010; Munneke et al. 2010; Beersen et al. 2011; van der Marck, Munneke, et al. 2013; Wensing et al. 2011; Canoy et al. 2015; van der Eijk, Bloem, et al. 2015; Sturkenboom et al. 2015; Sturkenboom et al. 2014; Ketelaar et al. 2013; Ypinga et al. 2018; Bloem et al. 2017). There is ample evidence that ParkinsonNet leads to a better quality of care, as reflected by a greater knowledge of PD among providers (Nijkrake et al. 2010a), better adherence to guidelines (Nijkrake et al. 2010; Munneke et al. 2010; Beersen et al. 2011), and a higher caseload per provider (Nijkrake et al. 2010; Munneke et al. 2010).

There is also evidence that ParkinsonNet enhances sustainability. Studies report better health outcomes and fewer PD-related complications (Beersen et al. 2011; Sturkenboom et al. 2014; Ypinga et al. 2018); and thereby reduced healthcare costs (Munneke et al. 2010; Beersen et al. 2011; Ypinga et al. 2018). Costs are also reduced by the greater efficiency of care, because therapists can do with fewer treatment sessions (Ypinga et al. 2018). Following the successful implementation process in the Netherlands, and stimulated by the demonstrated cost-effectiveness, other countries have also expressed an interest in introducing a similar type of network care. In the past few years, elements of the Dutch ParkinsonNet approach have successfully been introduced in the USA (ParkinsonNet 2014), Norway (ParkinsonNet 2016), Luxembourg, Germany and Czech Republic (Gal et al. 2017).

1.5 AIMS OF THIS THESIS

In this thesis, we seek to unravel certain important elements for seamless and sustainable care. We first analyse the current situation from the perspective of the person with PD, and identify the actual patients' needs. We then investigate potential innovations. Given that the organisation of PD care requires improvements in specific areas (see paragraph 1.3), we defined the research questions as presented in Table 1.3.

TABLE 1.3 | **General research question and sub questions**

General research question:	**How can we achieve seamless and sustainable care for people with PD?**
Sub questions:	• *What does the current patient journey of people with PD look like?* (chapter 2) • *What are the current seams in the care for people with PD?* (chapter 3) • *Can outcome-based payment models contribute to seamless and sustainable care for people with PD?* (chapter 4) • *Can the density of provider networks contribute to seamless and sustainable care for people with PD?* (chapter 5) • *How can we organise seamless and sustainable care for people with PD at the patient's home?* (chapter 6)

We will answer these questions in the following chapters:

- When aiming for seamless and sustainable care, it is important to know what care people with PD use. In **chapter 2** we will reconstruct the sex-specific 'journey' travelled by a person with PD through the healthcare system during the first five years of the disease course, based on claims data.
- In **chapter 3** we searched for elements that are missing in the care from the perspective of the person with PD, using the Voice of the Customer interview method.
- In **chapter 4** we present an overview of payment models in healthcare that include a financial incentive based on outcomes of care (outcome-based payment models, OMPMs) and their effects on the quality and costs. Since there are no experiments with OBPMs in specific PD situations, we made a comprehensive systematic review of all the literature about OBPMs in healthcare.
- **Chapter 5** analyses the density of provider networks and its effect on health outcomes and costs. This can be seen as an indication of volume efficiencies for chronic care.
- In **chapter 6** we discuss how recent innovations can bring the care for people with PD closer to home.
- The results of the previous chapters are discussed in **chapter 7**, which also gives the answers to our research questions.

CHAPTER 2

Sex-specific patient journeys in early Parkinson's disease in the Netherlands

F.P. Vlaanderen, Y. de Man, J.H. Krijthe, M.A.C. Tanke, A.S. Groenewoud, P.P.T. Jeurissen, S. Oertelt-Prigione, M. Munneke, B.R. Bloem and M.J. Meinders, 'Sex-Specific Patient Journeys in Early Parkinson's Disease in the Netherlands'. *Frontiers in Neurology*, 2019; 10:794.

ABSTRACT

Objective: To reconstruct a sex-specific patient journey for Dutch people with Parkinson's disease (PD) during the first five years after diagnosis.

Method: We analyzed a national administrative medical claims database containing data of all patients newly diagnosed with PD between 2012 and 2016 in the Netherlands. We performed time-to-event analysis to identify the moments when patients received care from neurologists, allied healthcare therapists or general practitioners. We also extracted relevant clinical milestones: unexpected hospitalization for PD, pneumonia, orthopedic injuries, nursing home admission and death. Using these data, we constructed the patient journey stratified for sex.

Results: We included claims data of 13,518 men and 8,775 women with newly diagnosed PD in the Netherlands. While we found little difference in neurologist consultations, women visited general practitioners and physiotherapists significantly earlier and more often (all p-values <0.001). After five years, 37.9% (n=3,326) of women had visited an occupational therapist and 18.5% (n=1,623) a speech & language therapist at least once. This was 33.1% (n=4,474) and 23.7% (n=3,204) for men. Approximately two years after diagnosis, PD-related complications (pneumonia, orthopedic injuries and PD-related hospitalization) occurred for the first time (women: 1.8 years; men: 2.3 years), and after five years, 72.9% (n=6,397) of women and 68.7% (n=9,287) of men had experienced at least one.

Discussion: Considering the strengths and limitations of our methods, our findings suggest that women experience complications and access most healthcare services sooner after diagnosis and more frequently than men. The identified sex differences extend the debate about phenotypical differences in PD between men and women.

2.1 INTRODUCTION

During the course of the disease, a patient with Parkinson's disease (PD) visits many different healthcare providers from different disciplines.(Radder et al. 2018) This 'journey through the healthcare system' varies per individual because of heterogeneity of symptoms, differences in disease progression rate, and the occurrence of PD-related complications. One important source of this variation might be sex differences in the presentation of PD. (Georgiev et al. 2017) For example, numerous studies confirm that the incidence and prevalence of PD is higher in men,(Baldereschi et al. 2000; Alves et al. 2009; Kovács et al. 2016; Georgiev et al. 2017; Marras et al. 2018) that the disease starts at an earlier age in men (Haaxma et al. 2007; Georgiev et al. 2017) and that the disease progresses faster in men.(Haaxma et al. 2007; Scott et al. 2000) In women, PD tends to be more often tremor-dominant,(Georgiev et al. 2017; Haaxma et al. 2007; Solla et al. 2012) while in men it is more often the akinetic-rigid type. (Georgiev et al. 2017; Baba et al. 2005; Picillo et al. 2017)

We do not know if these sex differences translate to different patient journeys between men and women with PD. But when striving for optimal patient-centered and integrated care, it is vital to understand what the patient journeys look like. As shown for other diseases(Kuo et al. 2015; Mehta et al. 2017; Hibbard, Mahoney, and Sonet 2017; Laveau et al. 2017) such insights can be used to improve access and optimize coordination of care. In this paper, we use medical claims data to reconstruct the sex-specific journey for Dutch people with PD during the first five years after diagnosis.

In the Netherlands, the patient's journey starts when a general practitioner makes a referral to a neurologist when symptoms of Parkinson's appear. Neurologists, all located in hospitals, make the diagnosis. Thereafter, a person with PD visits the hospital every three months, to see their neurologist, who is supported by nurses or nurse specialists. The nurse and neurologist work in close collaboration with allied healthcare professionals in the community, including, e.g., physiotherapists, occupational therapists and speech & language therapists. Hospital care is covered by the compulsory health insurance, whereas allied healthcare services are covered by additional insurance package, which is not compulsory but taken up by over 80% of the Dutch people. In addition, the Netherlands stands out with comparatively low out-of-pocket payments.(Maarse, Jeurissen, and Ruwaard 2016) This probably reduces any possible selection bias due to differences in price responsiveness among people with PD. In the analysis, we therefore focus on the most frequently involved healthcare disciplines (neurologists, allied healthcare therapists and general practitioners) and on recognized clinical milestones (PD-related complications, nursing home admission and death).

2.2 MATERIALS AND METHODS

To reconstruct the PD patient journey, we used medical claims data of all people with PD diagnosed between 2012 and 2016 in the Netherlands. The dataset was made available through Vektis, a not-for-profit organization that combines claims data of all Dutch healthcare insurance companies.(Vektis 2018) Since all Dutch citizens are obliged by law to have a healthcare insurance, the Vektis database contains the claims data of approximately 99.8% of the Dutch population(Zorgwijzer 2019) (17.3 million people (CBS 2018)). The claims database contains data on primary care, emergency care and hospital care, plus nursing home residency. The dataset was anonymized by Vektis, making available only the sex, year of birth, and a unique random identifier for each individual. The key to the identifiers was not available to the researchers.

Similar to a recent paper using similar Dutch claims data in PD,(Ypinga et al. 2018) we included only patients who had at least one diagnosis-related group code (DRG code) for PD. In the Netherlands, PD can only be diagnosed by a neurologist. We therefore regarded the first PD-related neurology DRG as the moment of diagnosis and, as such, as the starting point of the journey.

To reconstruct the patient journey, we included the professionals most frequently involved in the treatment of PD. These are neurologists (together with specialized PD nurses, since both claim their activities under the DRG code of the hospital), physiotherapists, occupational therapists, speech & language therapists, and general practitioners. For every included claim, we calculated how many days after the first diagnosis the activity had occurred. Next, we selected the 1st, 10th, 20th, and 30th visit to the general practitioner and the allied healthcare therapists. Unlike these disciplines, claims related to hospital care are defined in a DRG model, rather than by a pay-per-visit model. The first PD-related DRG includes at least one visit to a neurologist, but the actual number of visits may be higher. A subsequent PD-DRG can only start 90 days after the initial PD-DRG, and contains at least one visit to a neurologist or specialized PD nurse. Third and subsequent PD-DRGs can only start 365 days after the previous one. Consequently, the maximum number of PD-DRGs within the first five years after diagnosis is six. We therefore selected the first six PD-DRGs to assess utilization of neurologist. In a similar way, we identified the time after diagnosis until five clinical milestones in the patient's journey had been reached, using a methodology previously used in a comparable analysis:(Bjornestad et al. 2017) nursing home admission, hospitalization for three PD-related complications (unexpected hospitalization for PD, pneumonia, orthopedic injuries)(Ypinga et al. 2018; Muzerengi et al. 2016) and, finally, death.

We used event history analysis with Kaplan-Meyer estimators to determine after how many days the average patient received specific care or reached a clinical milestone.

This method deals with differences in length of follow-up between patients. The follow-up length was calculated for every patient as the number of days from diagnosis till death or till December 31st 2016, i.e., the last data point in the dataset. The median values of the time-to-event analysis were plotted on a timeline, representing the journey of the average person with PD. We constructed one timeline for men, and one for women. We chose median values over mean values because of the considerable differences in length of follow-up in our sample. Sex differences were statistically analyzed using log rank tests.

Standard Protocol Approvals, Registrations, and Patient Consents
This study was approved by the institutional review board of the Radboud University Medical Center with a waiver of consent for participants in the study.

Data Availability Policy
All data, published or not published within the article, is accessible through Vektis. Analyses were performed in SAS.

2.3 RESULTS

We included medical claims data of all 22,293 newly diagnosed patients in the analyses. As shown in Table 2.1, the population consists mainly of elderly individuals, a minority of whom lives in a nursing home before diagnosis.

TABLE 2.1 | General characteristics of the population (n=22,293)

	Men (n = 13,518; 60.6%)	Women (n = 8,775; 39.4%)
Age at diagnosis (mean, in years)	71.6 (SD: 9.9)	72.5 (SD: 10.2)
Early onset PD (<50 years at diagnosis, n (%))	421 (3.1%)	278 (3.2%)
Length of follow-up (mean)	2.5 years (SD: 1.4)	2.5 years (SD: 1.4)
Living in a nursing home at time of diagnosis (n (%))	673 (5.0%)	729 (8.3%)
Death during follow-up (n (%))	2469 (18.3%)	1284 (14.6%)

Healthcare utilization
Figure 2.1 shows two timelines representing the sex-specific patient journey of men and women with PD. Table 2.2 shows the inter quartile ranges (IQRs).

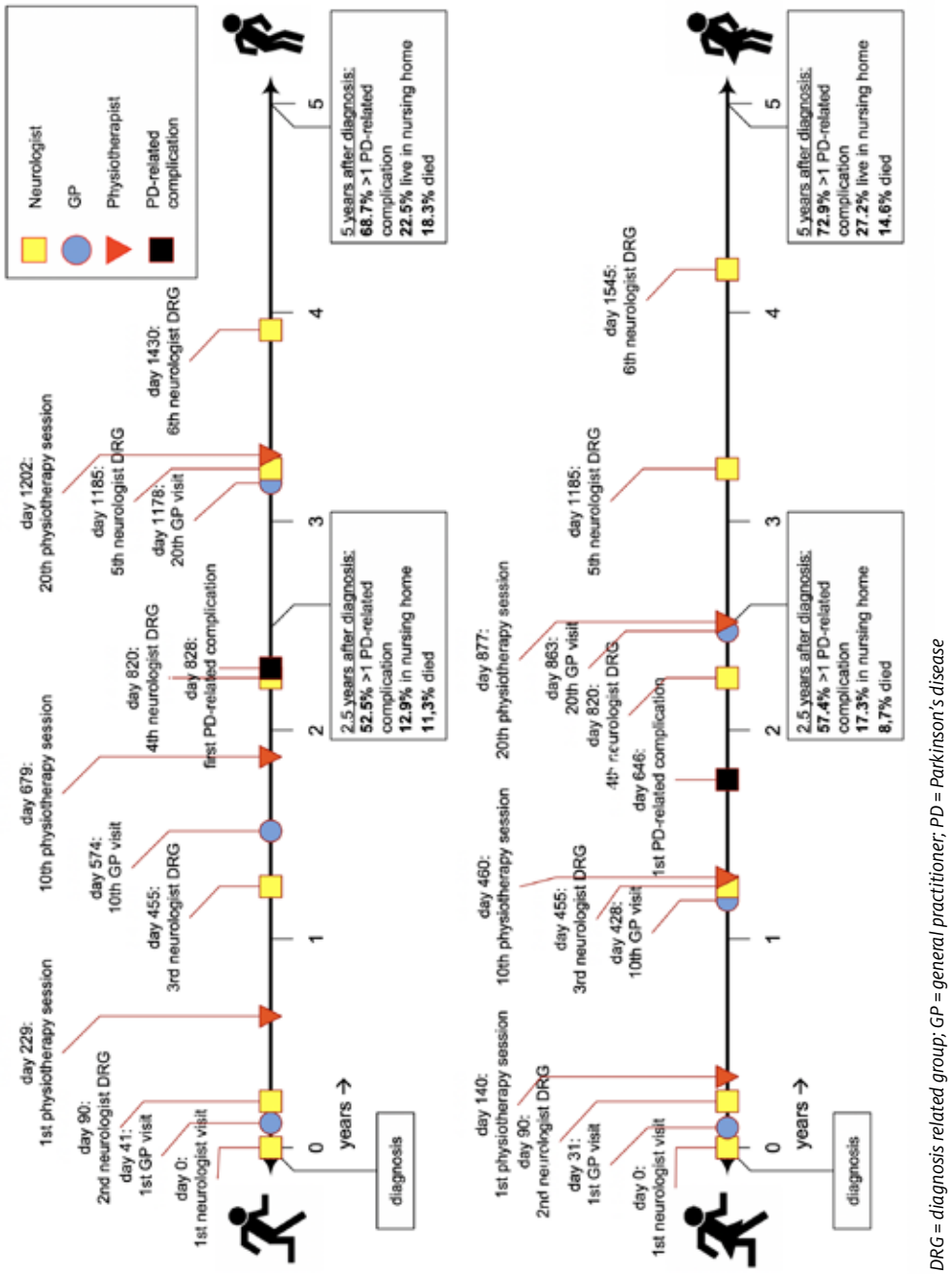

FIGURE 2.1 | **Timeline of the average Parkinson's patient journey during the first five years after diagnosis**

DRG = diagnosis related group; GP = general practitioner; PD = Parkinson's disease

TABLE 2.2 | **Time (in days, since diagnosis) till relevant provider contacts and clinical milestones during the patient journey, by sex**

	Women Median (1st – 3rd quartile)	**Men** Median (1st – 3rd quartile)	**P-value**
Provider contact			
1st neurology DRG	0 (0 – 0)	0 (0 – 0)	0.270
2nd neurology DRG	90 (90 – 240)	90 (90 – 210)	0.079
3rd neurology DRG	455 (360 – >1826)	455 (330 – 1454)	0.040
4th neurology DRG	820 (575 – >1826)	820 (575 – >1826)	0.280
5th neurology DRG	1185 (935 – >1826)	1185 (935 – >1826)	0.348
6th neurology DRG	1545 (1180 – >1826)	1430 (1180 – >1826)	0.333
1st general practitioner visit	31 (8 – 87)	41 (11 – 116)	<0.001
10th general practitioner visit	428 (231 – 824)	574 (301 – 1112)	<0.001
20th general practitioner visit	863 (505 – 1605)	1178 (659 – >1826)	<0.001
30th general practitioner visit	>1826 (830 – >1826)	>1826 (1082 – >1826)	<0.001
1st physiotherapist visit	140 (16 – 826)	229 (38 – 1303)	<0.001
10th physiotherapist visit	460 (136 – >1826)	679 (177 – >1826)	<0.001
20th physiotherapist visit	877 (268 – >1826)	1202 (341 – >1826)	<0.001
30th physiotherapist visit	>1826 (415 – >1826)	>1826 (530 – >1826)	<0.001
1st occupational therapist visit	>1826 (986 – >1826)	>1826 (1247 – >1826)	<0.001
1st speech & language therapist visit	>1826 (>1826 – >1826)	>1826 (>1826– >1826)	<0.001
Clinical milestones			
Nursing home admission	>1826 (1571 – >1826)	>1826 (>1826 – >1826)	<0.001
1st PD-related complication			
pneumonia	>1826 (661 – >1826)	>1826 (686 – >1826)	0.879
orthopedic injuries	1114 (347 – >1826)	>1826 (527 – >1826)	<0.001
hospitalization	>1826 (>1826 – >1826)	>1826 (>1826 – >1826)	0.359
All PD-related complications	646 (179 – >1826)	828 (214 – >1826)	<0.001
Death	>1826 (>1826 – >1826)	>1826 (>1826 – >1826)	<0.001

Approximately one month after diagnosis, patients first visited their general practitioner (women after 31 days; men after 41). Thereafter, women saw their general practitioner approximately once every six weeks (43 days). Men saw their general practitioner less often: approximately once every eight to nine weeks (59 days). For both sexes the frequency declined after the 20th visit (median >5 years for 30th visit). In all analyses, women visited their general practitioner significantly earlier than men (p-values <0.001).

Three months after diagnosis, many patients saw their neurologist or specialized PD nurse again (both for men and women median = 90 days). However, a substantial part of the population also visited their neurologist or specialized PD nurse much later for the second time, i.e., not before eight to nine months after diagnosis (75th quartile value = 210 days (men) and 240 days (women)). After the second visit, the frequency of claimed neurology DRGs was about once a year for both sexes, i.e. they visited a neurologist or specialized PD nurse at least once a year. Except for the third visit, where men used neurologist services slightly earlier than women, no significant sex differences were found.

Women with PD started their physiotherapy treatment approximately five months after diagnosis (median = 140 days). Their first twenty physiotherapy sessions took place about once every five to six weeks. Men started to visit a physiotherapist later after the diagnosis then women: eight months after diagnosis (median = 229 days), and with a lower frequency: once every seven to eight weeks. For both sexes the frequency declined after the 20th session (median >5 years for 30th physiotherapy sessions). In all analyses, women used physiotherapist services significantly earlier and within a shorter timespan than men (p-values <0.001).

For occupational therapists and speech & language therapists, the median values for the first visits were not reached within the follow-up time of five years. Therefore, they are not displayed in Figure 2.1. After five years, 37.9% of the women (n=3,326) and 33.1% of the men (n=4,474) had visited an occupational therapist at least once. This was 18.5% for women (n=1,623) and 23.7% for men (n=3,204) for the first visit to speech & language therapist. These differences were statistically significant (p-values <0.001)

Clinical milestones

Approximately two years after diagnosis, the first PD-related complication occurred. For women the median value was 1.8 years (IQR= 0.5 - >5 years); for men this was 2.3 years (IQR= 0.6 - >5 years; p-value <0.001). We added the Kaplan-Meyer curve of this analysis in Figure 2.2. As shown in Table 2.2, orthopedic injuries were the most common complication, and occurred earlier in the course of the disease in women (p-value <0.001). Five years after diagnosis, the percentage of patients that had experienced at least one PD-related complication was 72.9% in women (n=6,397) and 68.7% in men (n=9,287). The percentage of women admitted to a nursing home rose from 8.3% (n=728) before diagnosis to 27.5% (n=2,413) after five years of PD. In men this increase is from 5.0% (n=676) to 22.5% (n=3,042) (p<0.001). During the first five years after diagnosis, significantly fewer women died (14.6%) than men (18.3%, p-value = <0.001).

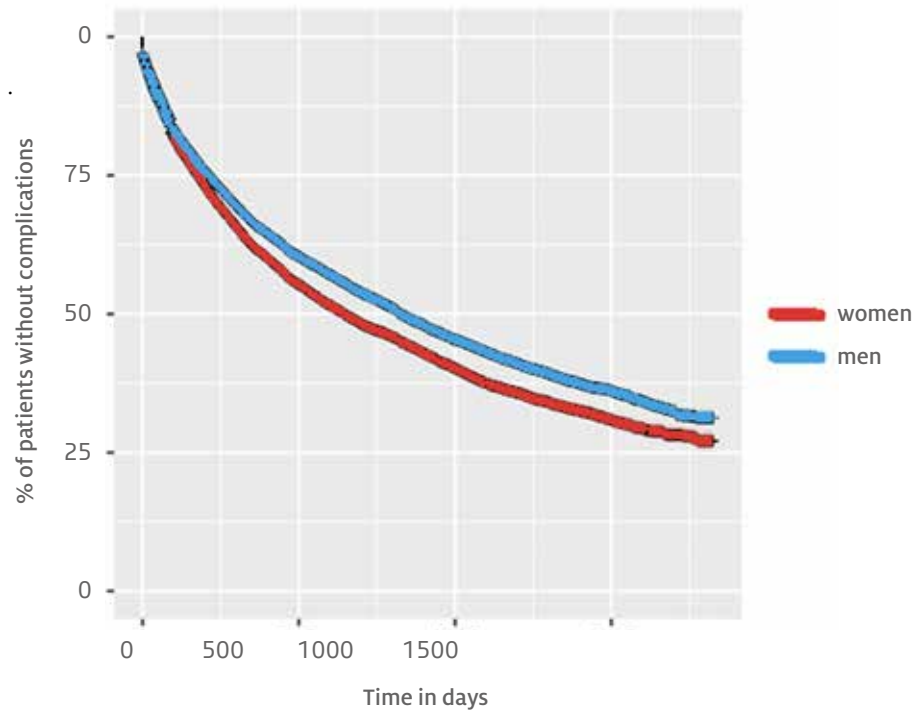

FIGURE 2.2 | **Kaplan-Meyer curve of the time-to-event analysis for PD-related complications**

2.4 DISCUSSION

The reconstruction of the Parkinson patient's journey through the Dutch healthcare sector during the first five years after diagnosis, reveals quantitative information about healthcare utilization and the occurrence of clinical milestones over time. It also reveals sex differences: in the Netherlands, women visit most of the included healthcare professionals sooner after diagnosis and more frequently. In addition, PD-related complications occur earlier in women than in men. A sizeable subgroup of patients is admitted to nursing homes within five years after diagnosis. Again, this happens more frequently in women. Finally, 14.6% of the women and 18.3% of the men died within five years after the diagnosis.

Relation with previous findings

Our findings confirm and extend earlier work, from both inside and outside the Netherlands. The characteristics of our population are comparable to earlier work when it comes to general incidence(Hijdra, Koudstaal, and Roos 2012) and the male predomination of the disease.(Baldereschi et al. 2000; Alves et al. 2009; Kovács et al.

2016; Georgiev et al. 2017; Marras et al. 2018) Comparable values for age at diagnosis and percentage of early-onset PD were also found before.(Van Den Eeden et al. 2003) However, while most studies found a faster disease progression in men,(Haaxma et al. 2007; Scott et al. 2000) our findings suggest a more rapid disease progression in women, since they visited their healthcare professionals sooner and experienced orthopedic injuries earlier and more often after diagnosis. However, there might be other explanations for this observation. First, the included women were living relatively more often in a nursing home before diagnosis, indicating that they were probably in a worse physical condition at the outset. This might make them more prone to develop complications and also explain the more intense healthcare utilization. Patients living in nursing homes might also have easier access to the in-house allied health therapists. Second, the findings can indicate a doctor or patient delay in the diagnostic process in women, meaning that women receive the diagnosis relatively late in the course of the disease. This has been found earlier.(Saunders-Pullman et al. 2011) Alternatively, women might find their way to healthcare professionals more effectively (or faster). This has been observed for other diseases(Addis and Mahalik 2003; Cohen 2009) but not previously for PD.

What surprised us was the high mortality rate and that patients experienced their first PD-related complication already two years after the diagnosis. No PD-specific literature is available to compare these results with. The average age at onset of 72 years is in line with other population-based cohort studies.(Darweesh et al. 2017; Savica et al. 2016; de Lau and Breteler 2006) The finding that 5.08.3% of the patients are living in a nursing home at diagnosis, which is are relatively high, can be understood when considering that the Netherlands has relatively one of the largest long term care sectors in the world.(Mosca et al. 2016) Given that patients living in a nursing home are likely more vulnerable, this might explain the mortality and complication rates.

Strengths and limitations

Our methods have strengths and limitations. An important strength is that our dataset contained all newly diagnosed patients in the country over a period of five consecutive years. This reduces a potential selection bias. However, some selection may have resulted from our inclusion criteria. Since we included all patients with a first PD DRG, there might be some cases where the initial diagnosis of PD was wrong. PD is hard to diagnose, with reported diagnostic error rates of >10%.(Skinner, Scott, and Martin 2016) Therefore, we cannot exclude that some patients had other conditions, in particular one of the forms of atypical parkinsonism.(Hijdra, Koudstaal, and Roos 2012) Our sample most likely included people who were incorrectly diagnosed. A review of the literature, including 11 studies, concluded that the validity of clinical diagnosis of PD Is not satisfying, which was the case for both non-experts and movement disorder specialists.(Rizzo et al. 2016) We were not able to correct for this error in our analysis.

However, since our population characteristics matched well with previous reports, we do not think that all these factors affected our data on a large scale. Moreover, it is unlikely that this diagnostic error affected men and women differentially. Another strength is that our study is based on highly standardized claims data. And we only used items that, although self-reported by healthcare professionals, are known to be reliably completed.(Vektis 2017)

As claims data don't include detailed information about the clinical status of the patients, we were not able to correct for factors that confound and/or modify the relationship between sex and complications.(Bloem et al. 2018) This holds particularly true for co-morbidity and PD-related complications that are more frequently associated to female sex (e.g., dyskinesia, motor and non-motor complications).(Picillo et al. 2017) Also, the sex-difference in the occurrence of orthopedic injuries might be explained by the female predominance in osteoporosis. Therefore, our findings require confirmation in other, independent datasets.

Finally, our findings might be difficult to extrapolate to other countries with another organization of the healthcare system. For example, duration of visits and treatment intensity can differ between countries. Moreover, the presence of ParkinsonNet in the Netherlands contributed to the quality and role of allied health care professionals, and stimulated access to specialized and multidisciplinary Parkinson care.(Bloem et al. 2017)

Practical implications

Comparable work on other diseases suggests that our reconstruction of the patient journey may lead to better patient-centered care delivery. Specifically, it provides healthcare professionals an overview of where and when particular physicians get involved, which might reveal errors in providers' perspectives.(Tan et al. 2017) It might act as a useful tool to gain insight in patient experiences,(Tan et al. 2017; Kuo et al. 2015) to reveal barriers to access,(Kelly et al. 2017; Mehta et al. 2017) to detect gaps in care delivery,(Thrift-Perry et al. 2018; Roughead, Semple, and Rosenfeld 2016) and to improve coordination and quality of care.(Kelly et al. 2017; Crepaz and Curry 2013) The identified sex differences might contribute to the debate about differences in PD between men and women, extending earlier work on different phenotypes to now include contrasts in healthcare utilization as well. We hope these insights can lead to better and more personalized care for people with PD of both sexes.

CHAPTER 3

The voice of the Parkinson customer

F.P. Vlaanderen, L. Rompen, M. Munneke, M. Stoffer, B. Bloem, M. Meinders,
'The voice of the Parkinson customer'. *Journal of Parkinson's Disease*. 2019; 9(1):197-201.

3.1 INTRODUCTION

To improve the care for patients with chronic neurological conditions like Parkinson's disease, identifying the core needs of patients is crucial (Kleiner-Fisman, Gryfe, and Naglie 2013). Usually, these needs are assessed by interviews, focus groups or questionnaires among patients or healthcare professionals. Kaiser Permanente, an American not-for-profit health plan, developed the Voice of the Customer (VoC) approach to assess the needs of their members from a person-centered perspective. This method, which was borrowed from the field of industry to probe the clients' needs, applies sets of qualitative research methods (including semi-structured interviews, video ethnography and participatory observations) to identify the needs of end-users, i.e. patients, family, clinicians and staff. Kaiser Permanente used these methods successfully to develop quality improvement programs and to improve existing programs to better meet people's needs and deliver better outcomes (Neuwirth et al. 2012; Kaiser Permanente 2016).

In June 2015, we applied the VoC approach for the first time outside of the Kaiser Permanente system, and also for the first time in the field of Parkinson's disease. The VoC approach, as described in this paper, aimed to identify the needs of Dutch patients with Parkinson's disease, in order to further optimize the care they receive.

3.2 METHODS

The VoC approach consisted of three steps: (1) capturing patient needs by means of semi-structured interviews with patients, relatives and healthcare providers in their private environment; (2) preparing a comprehensive summary of the contents discussed in the interviews; and (3) prioritizing needs in a consensus meeting, in which all parties participate.

For the first step, all Parkinson patients visiting one of the two hospitals in Nijmegen, the Netherlands, were identified using the hospital information systems. There were two inclusion criteria. First, patients needed to be diagnosed with Parkinson's disease. For this purpose, the diagnosis in all participants was verified by a neurologist with experience in movement disorders, based on accepted international criteria. Second, patients had to live in one of three representative suburbs of the city of Nijmegen, the Netherlands. All identified patients fulfilling the inclusion criteria then received a recruitment letter with information about the VoC approach.

Participants completed the informed consent form. They were asked which healthcare professionals they normally visited. All identified healthcare professionals who were working in the three suburbs were then invited for an interview. To adequately capture

the voice of the patient, we aimed for inclusion of 24 patients and 12 healthcare professionals. These numbers were based on the prior experiences of Kaiser Permanente, where answers saturated when these sample sizes were used.

The recruited patients, sometimes accompanied or represented by their relatives, and the healthcare professionals were subjected to a semi-structured interview of approximately one hour. Interviews were always performed by two interviewers. During the interviews, the interviewers positioned the interviewee in an expert role. The interviewers posed questions, listened and observed, while creating an atmosphere that facilitated an open conversation and avoided socially acceptable answers. Interviewers were instructed to probe for clarification and examples when desirable. All respondents were asked about their positive and negative experiences with current care, and their opinion on which aspects needed improvement. The interviewed healthcare professionals were asked to answer these questions, both from their own perspective and from the perspective of their patient. The interviews took place at the home of the patient or the practice of the healthcare professional, and all were recorded on video.

To maintain focus and accelerate the analysis, all interviews were scheduled within one week. This required a relatively large pool of 12 interviewers. To diminish inter-rater bias, we developed specific interview guides for interviews with patients, relatives of patients and healthcare professionals. These guides were based on the experiences of Kaiser Permanente. Besides instructions for the interviewers, the guides did not contain a topic list. This was left out to let the interviews proceed as open as possible. They were presented and discussed in a training session for the interviewers. Subsequently, the interviewers carried out a test interview, to complement the preparation phase.

For the second step, the interviewers translated the completed interviews into patient needs. At the end of each day of the interview week, the interviewers came together to review their videos and discuss their findings. During these meetings, the interviewers drew conclusions as to the most frequently expressed patient needs. After the interview period, the interviewers made a comprehensive video that showed the main conclusions, combined with illustrative quotations by the interviewees. This video formed a comprehensive outline of the patient's story, and aimed at serving as a tool to generate ideas for future improvement projects.

The third step was to prioritize the identified patient needs. During a consensus meeting with the interviewed patients and healthcare professionals, the most frequently expressed patient needs were presented on posters. After discussing the posters and viewing the comprehensive video, the patients and relatives prioritized

the patient needs by allocating a maximum of three points to each of the different needs. They could allocate all their points to one need or divide these between two or three needs. The total number of points represented the importance of the topic.

The study was not presented to the institutional review board for ethical approval. The Voice of the Customer approach was part of a larger quality improvement program for Parkinson patients, for which in the Netherlands no approval by an institutional review board is required. Even though this was not formally required, we did obtain our own informed consent from every participant, after the purpose of the project and the patients' contribution was carefully explained.

3.3 RESULTS

In total, 89 patients were invited, of whom 23 agreed to participate (26% participation rate). Twenty of them were interviewed, of which nine had an interview together with their spouse. For three other patients, a close relative participated in the interview instead. Additionally, 81 healthcare professionals were invited for an interview. Eleven healthcare professionals were included (14% participation rate), leading to a total number of 34 interviews. Each interview lasted approximately 70 minutes. See Table 3.1 for more detailed information about the respondents.

TABLE 3.1 | Overview of interviewed individuals

N	Patients (n=20) and relatives (n=12)
20	Patients, of whom nine were accompanied by their relative during the interview
1	Spouse of patient
1	Son of patient
1	Friend of patient
	Healthcare professionals (n=11)
2	Specialized Parkinson nurse
2	Social worker
1	Physiotherapist
1	Occupational therapist
1	Speech and language therapist
1	General practitioner
1	Elderly care physician
1	Home care worker
1	Neuro-psychologist

TABLE 3.2 | **Top 10 priorities of patient needs, as retrieved by the VoC approach.**

	Item	Points	Illustrative quotes (translated to English)
1	Desire for self-management	22	"The three of us met: the neurologist, the nurse and I. They were talking to each other using technical terms, while I was just sitting there. I became a bit angry: they were talking about me, so I would like to understand what they were saying." "Going to the hospital was compulsory; not because we felt the need to go at that moment." "I want to decide myself if I need care. That is what I call self-management."
2	Better collaboration between my different healthcare professionals	21	"I am seeing a cardiologist. I noticed this was not communicated to my neurologist. That makes me question: 'This is about my life, and two people in the hospital are individually working on it. Why are they not working together?'" "I think specialists are named 'specialists' because they work in their own work field, of which they know almost everything, but they are very isolated from each other."
3	More time for discussing the future	20	"I fear the future. I notice that I am thinking a lot about how the disease will progress: how will it be? How will my life look like?" "I have it for five years now. The neurologist told me: 'up to now it seems that the disease progresses quite slowly. We can expect this trend to continue in the future.' I feel the need to know this type of information."
4	One healthcare provider who can act as a personal case manager	18	"Sometimes I have panic attacks. It helps if I can contact the nurse to talk about it. But if I call and I get an appointment in two weeks, it is useless. Then I do not need it anymore." "I have a good relationship with the specialized nurse. She knows me quite well by now. I call this 'warm care'. She has patience for me, and she recognizes me when we coincidently encounter outside the hospital." "It would certainly help if there was somebody who said: 'I want to help you during the whole process. You will encounter this and that.' This makes you feel prepared."
5	More knowledge of the disease	11	"Often home care nurses lack specific knowledge of Parkinson's." "Pharmacists lack detailed knowledge of the different types of medication."
6	More support from my pharmacist	10	"There are three different brands of Madopar, so things sometimes go wrong when ordering medication. The attitude of pharmacists regarding this is frustrating."
7	Increased focus on the needs of my spouse	9	"You do not have Parkinson's alone. The whole family has Parkinson's. It would be nice if this issue was given some attention."
8	More contact with other patients	7	"I strongly feel the need to talk to people in the same situation." "That is the best that can happen to you: you encounter someone who also has the disease. Then you do not need many words to explain what is going on."
9	More provision of information	6	"If I look back, I think it would have been useful to be explained things straight away. Like, 'okay, your father is diagnosed with Parkinson's disease. Let's sit down together with all the involved healthcare providers and explain what this will mean for you as a family member of a Parkinson patient.'"
10	Less fragmentation of healthcare	6	"It would be nice if I could see the same neurologist every time; the same nurse, the same therapist. They should then sit together once in a while and discuss my case as their mutual patient. If they had more contact with each other and did this, they would know who I am."

The interviewers extracted the 10 most frequently expressed patient needs from the interviews, which were subsequently prioritized during a consensus meeting. Patients were generally more concerned about the impact of Parkinson's disease on their daily lives than about the bio-medical aspects of their disease. Their top unmet needs were: (1) more self management; (2) better interdisciplinary collaboration between different healthcare professionals; (3) more time to discuss the future and possible scenarios; and (4) one healthcare professional as a single point of access, acting as a personal case manager, either to solve problems directly or to direct patients to the professional best equipped to address the problem at hand (see Table 3.2).

3.4 DISCUSSION

The VoC approach is an innovative, person-centered and relatively fast way to reveal the most important needs of Parkinson patients. We found that the most urgent patient needs, report by themselves, concern the social, emotional or domestic domain of the patient.

Some of our findings confirm the results of previous research: a large European survey among 2068 patients revealed that only 11.6% of patients feels involved in treatment decisions(Bloem and Stocchi 2015). This is in line with the desire for more self-management, similar to what we found, which includes the involvement in decision making. However, most other studies found different needs: the main results of the previously mentioned European survey found unmet needs in the diagnostic process(Bloem and Stocchi 2012), the diagnosis delivery(Bloem and Stocchi 2012) and access to allied healthcare professionals and specialized nurses(Stocchi and Bloem 2013). Other studies reported a lack of information provision, emotional support, and multidisciplinary collaboration of healthcare professionals(van der Eijk, Faber, Al Shammaa, et al. 2011; Buetow et al. 2008). The differences in identified patient needs may be due to the fact that most of the existing literature consists of the standard type of questionnaires that focus on the biomedical domain or the professional-patient relation. In contrast, the VoC method is a deep interviewing method that really places the patient in the expert role. This may have led to an accentuation of the social, emotional or domestic domain. For example, a key issue such as the desire to have one easily approachable healthcare provider who can act as a personal case manager, has not been described before. Sharing such personal matters with a stranger (an interviewer whom patients did not know) requires an open dialogue and an environment in which the interviewee feels sufficiently safe to share his or her deepest feelings. This might be the area where the VoC approach can add the greatest value, compared to more traditional methodologies like surveys or focus groups.

An important limitation of our research is the low participation rate of invited patients (26%) and healthcare professionals (14%), which might lead to inclusion bias. Additionally, no neurologist or movement specialist participated in the interviews. Since the VoC approach was performed as a quality improvement program without the primary intention to conduct research, no data about those who did not participate were collected.

The VoC approach used here revealed that the perception of "being healthy" is broader than simply achieving an acceptable level of symptoms. It also includes the ability to maintain a social life and participate in society. These new results should now be used to further optimize the care for patients with Parkinson's disease.

CHAPTER 4

Design and effects of outcome-based payment models in healthcare: a systematic review

F.P. Vlaanderen, M.A. Tanke, B.R. Bloem, M.J. Meinders, F. Eijkenaar, F.T. Schut en P.P.T. Jeurissen, 'Design and effects of outcome-based payment models: a systematic review'. *European Journal of Health Economics*, 2019;20(2):217-232.

ABSTRACT

Introduction: Outcome-based payment models (OBPMs) might solve the shortcomings of fee-for-service or diagnostic related group (DRG) models by using financial incentives based on outcome indicators of the provided care. This review provides an analysis of the characteristics and effectiveness of OBPMs, in order to determine which models lead to favourable effects.

Methods: We first developed a definition for OBPMs. Next, we searched four data sources to identify the models: 1) scientific literature databases; 2) websites of relevant governmental and scientific agencies; 3) the reference lists of included articles; and 4) experts in the field. We only selected studies that examined the impact of the payment model on quality and/or costs. A narrative evidence synthesis was used to link specific design features to effects on quality of care or healthcare costs.

Results: We included 88 articles, describing 12 OBPMs. We identified two groups of models based on differences in design features: narrow OBPMs (financial incentives based on quality indicators) and broad OBPMs (combination of global budgets, risk sharing, and financial incentives based on quality indicators). Most (5 out of 9) of the narrow OBPMs showed positive effects on quality, the others had mixed (2) or negative (2) effects. The effects of narrow OBPMs on healthcare utilization or costs, however, were unfavourable (3) or unknown (6). All broad OBPMs (3) showed positive effects on quality of care, while reducing healthcare cost growth.

Discussion: Although strong empirical evidence on the effects of OBPMs on healthcare quality, utilization, and costs is limited, our findings suggest that broad OBPMs may be preferred over narrow OBPMs.

4.1 INTRODUCTION

In most developed countries, policy makers are searching for payment systems which stimulate the quality of care and reduce healthcare costs. The predominant fee-for-service and diagnosis related group (DRG) models incentivize volume, and are therefore widely considered to be an important reason for rising costs in healthcare (Orszag and Ellis 2007). While incentivizing volume can lead to reduced waiting times and better access to healthcare, fee-for-service and DRG models lack incentives for improving quality: providers are paid for the quantity of care they deliver, not for the impact on the health status of their patients (Tai, Kalanithi, and Milstein 2014). Since the start of this century, pay-for-performance (P4P) models became popular as a response. In P4P models, reimbursement of healthcare providers explicitly depends on meeting predefined quality targets, which to date have largely been based on process and structure indicators (Nicholson et al. 2008). Though models based on these indicators have been studied extensively, evidence that these P4P models are (cost-) effective is limited (Eijkenaar et al. 2013; Milstein and Schreyoegg 2016). Additionally, it is still unclear whether the results of initially effective P4P models are sustainable (Eijkenaar et al. 2013; Milstein and Schreyoegg 2016; Ryan, Nallamothu, and Dimick 2012). Many authors emphasize the important influence of adequate design features, including the selection of incentivised indicators, on the effectiveness of P4P models (Roland and Campbell 2014; Conrad and Perry 2009; Eijkenaar et al. 2013; Eijkenaar and Schut 2015; Eijkenaar 2013; Jha 2013; Mehrotra, Sorbero, and Damberg 2010; Rosenthal and Dudley 2007; Werner and Dudley 2009; Roland 2012).

Over the last decade, the different shortcomings of P4P models based on structure and process indicators have been addressed by an increased incorporation of outcome indicators. The question is if this increased focus on outcomes has resulted in better quality of care and/or reduced cost growth, or if there are other design features that are (more) important.

However, a comparative evaluation of payment models with an increased focus on outcomes is lacking. Therefore, we conducted a systematic review of the literature on the effects of these new models. Our objective is to synthesize the evidence of the effects on quality of care, healthcare utilization and healthcare costs. This will lead to better understanding of the consequences of these models, and will help to determine which design features lead to favourable effects, and why. In addition, it might lead to further development and implementation of effective payment models.

In this paper, we use the term ‘outcome-based payment models’ (OBPMs) to denote payment models with a substantial reliance on outcome indicators. Although this term is frequently used in the literature, there is no uniform definition (Eijkenaar, van

de Ven, and Schut 2012; Hayen et al. 2013). For example, there is no standard about the minimum use of outcome indicators, while only a few models use outcome indicators exclusively. When creating a definition for OBPMs, we noted that in P4P models outcome indicators typically contribute less than 10% to the performance-related incentive payments (see the examples in (Eijkenaar, van de Ven, and Schut 2012; Eijkenaar and Schut 2015; Hayen et al. 2013; Eijkenaar 2012)). Based on this finding and on expert opinions in the field (Appendix 4.3), we choose for a pragmatic approach to consider programmes OBPMs if at least 10% of the performance -related incentive payment is determined by scores on outcome indicators. We adopted the following definition:

An outcome-based payment model is a payment model in healthcare in which the performance-related incentive payments for the healthcare providers depend for at least 10% on outcomes of the provided care, and which is designed to stimulate favourable effects in terms of quality of care or healthcare costs.

We address the following questions: 1) What are the design features of OBPMs and to what extent do they differ from each other? 2) What are the effects of OBPMs on quality of care, healthcare utilization, and healthcare costs?

4.2 METHODS

Inclusion and exclusion criteria

Included articles had to describe the effects on quality of care, healthcare utilization or healthcare costs of at least one OBPM that matched the definition mentioned in the introduction. In this article quality of care is assessed by the scores on quality indicators according to the donabedian framework (structure, process and outcome indicators) (Donabedian 1988). 'Outcome' is defined as *'the effects of care on the health status of patients and populations'* (Donabedian 1988). We do not distinguish between intermediate outcomes (e.g. blood pressure values), final outcomes (e.g. mortality, complication rates, and hospital readmissions) and patient-reported outcomes. 'Healthcare costs' are defined according to the definition of the OECD: *'the sum of expenditure on activities that – through application of medical, pharmaceutical, and nursing knowledge and technology – have certain healthcare related objectives'* ((OECD) 2014).

Articles written in English and published between January 2000 and October 2016 were included. We only included effects that were achieved in OECD countries ((OECD) 2011), since the aims and contexts of programmes in other countries are too different to allow a useful comparison. To be as comprehensive as possible, we did not focus on a specific healthcare sector (e.g. in- or outpatient care), despite typical differences

in incentive structures that might exist across sectors. There was also no restriction in study design; qualitative studies, quantitative studies, and reviews were all eligible for inclusion. However, articles describing only simulated or expected effects were excluded. Because we expected that many evaluations of OBPMs are not published in scientific peer-reviewed journals, we included governmental and other research reports (provided that they matched our inclusion and exclusion criteria) to ensure a complete inclusion of information. Letters, editorials and viewpoints that did not contain primary research were excluded.

Search strategy

We used four data sources to ensure a comprehensive search. Firstly, we searched three databases with scientific literature (Medline, the Cochrane Library, and EMBASE), using the keywords listed in Appendix 4.1. Secondly, we consulted websites of relevant governmental and/or scientific agencies (see Appendix 4.2). Thirdly, we searched through the references of the yielded documents. Finally, we consulted several experts in the field, all of whom responded (see Appendix 4.3).

Selection procedure

Titles and abstracts of the documents yielded by the three scientific databases were checked for duplicates and remaining articles were screened for relevance. Full-texts of seemingly relevant articles were subjected to the inclusion and exclusion criteria. To determine if a model matched our definition of an OBPM, we sometimes searched for additional information about the model on the Internet via Google, using programme-specific keywords. The selection procedure was done independently by two reviewers. Meetings were held to minimise interobserver bias. Differences were resolved in a discussion between the reviewers, if necessary after consultation of a third reviewer.

Next, articles found on websites of the consulted agencies, articles that were brought to our attention by the consulted experts, and articles retrieved from references of included documents were subjected to the inclusion and exclusion criteria.

Data extraction

To extract and summarize the data, we developed an extraction form. This form contained the three elements:

- Name, country, and period in which the model was operating.
- Design features of the payment model
- Effects on quality of care, healthcare utilization and healthcare costs.

A methodological challenge was the fact that payment models tend to change over time, sometimes on an annual basis, e.g. indicators were added or removed, payment structure changed. To address this, we searched for additional information about

the changes in programme design over time. If due to these changes the model did not meet our definition of OBPM in a specific year, the results achieved in that year were not taken into account. The process of data extraction was performed by two independent reviewers.

Study appraisal

To appraise the methodological quality of the included quantitative studies, we used the generic and widely applied method described by Downs & Black (Downs and Black 1998). In the Downs & Black method, articles receive points on 27 items covering four domains: reporting, external validity, internal validity and power. The more points an article receives, the higher the methodological quality of the article. The maximum number of points is 32 (Downs and Black 1998). We chose this generic appraisal method because of the expected heterogeneity of the included study designs, e.g. interrupted time series, observational cohort studies, and cross-sectional studies. To determine the methodological quality of included qualitative studies and reviews, we used the Critical Appraisal Skills Programme checklists ((CASP) 201787). These appraisal methods have been used in other systematic reviews of the effects of payment models in healthcare (van Herck et al. 2010; Gillam, Siriwardena, and Steel 2012; Eijkenaar et al. 2013).

The study appraisal was performed by one reviewer; a second reviewer then did an independent review of all qualitative studies and reviews, plus a random selection of 10% of the included quantitative studies. Meetings were held to minimise inter-observer bias. Differences were resolved in a discussion between the reviewers, if necessary after consultation of a third reviewer.

4.3 RESULTS

Included studies

Figure 4.1 summarizes the search flow. The 88 included articles contained 75 quantitative studies, 8 qualitative studies, 3 research reports, and 2 reviews. All quantitative studies had a quasi-experimental design (difference-in-difference and case-control design). They had an average Downs & Black score of 11.7 (out of 32) and a standard deviation of 1.9 (Appendix 4.4). Most points were lost on items about internal validity and statistical power.

One quantitative study contained results for two OBPMs, and one policy report contained results of three OBPMs. The rest of the yielded documents described only one model. In total, we identified 29 OBPMs (Appendix 4.5), of which 12 could be included for our analysis. Table 4.1 and Table 4.2 provide the general characteristics and the design features of the 12 included OBPMs.

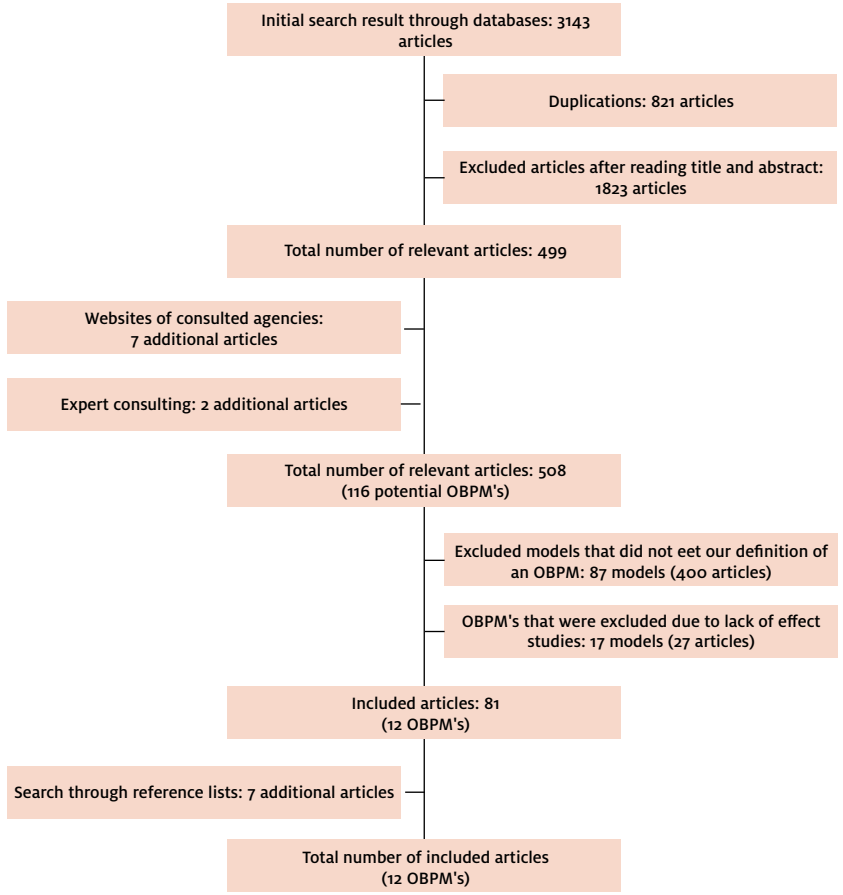

FIGURE 4.1 | **Search flow and results**

Based on the general characteristics (Table 4.1) and the design features (Table 4.2), we identified two types of OBPMs. We called the first group 'narrow OBPMs'. The models comprising this group focus exclusively on explicit financial incentives for objectively measured quality, with the incorporation of relatively many outcome indicators (i.e. pertaining to >10% of performance-related reimbursement). In these models, providers earn bonuses and/or suffer penalties based on their scores on a predefined set of indicators. These models typically target one provider type (e.g. hospitals, primary care physicians) and/or specific clinical areas (e.g. care for acute myocardial infarction). The other group of models is called 'broad OBPMs'. These models encompass the entire provider payment by combining global budgets and shared savings incentives with explicit financial incentives for quality indicator scores. This group of models generally targets multidisciplinary provider groups providing different types of care for their patient population.

TABLE 4.1 | **Characteristics of the 12 included outcome-based payment models**

Name, country, period, and references	Healthcare purchaser	Targeted care	Targeted healthcare providers	Outcome indicators and their contribution (in %) to the performance-related payment size:
Alternative Quality Contract (AQC) USA; Since 2009 ***(Song et al. 2011; Song et al. 2012; Eijkenaar and Schut 2015; Eijkenaar, van de Ven, and Schut 2012)***	Blue Cross Blue Shield (BCBS) private; HMO	All care for BCBS insured	Integrated care model: all providers involved in targeted care	Cholesterol levels; HbA1c levels; blood pressure (35,3%)*
Commissioning for Quality & Innovation (CQUIN) UK Since 2010 ***(Eijkenaar and Schut 2015; McDonald et al. 2013)***	National Health Service (NHS) public; single purchaser	Acute care, ambulance service, mental health care, and home care for NHS	Multiple provider model: all providers involved in targeted care	Unknown: differs locally (usually >10%)
Hospital Quality Incentive Demonstration (HQID) USA; 2003-2009 ***(Bhattacharyya, Mehta, and Freiberg 2008; Epstein, Jha, and Orav 2014; Eijkenaar and Schut 2015; Eijkenaar, van de Ven, and Schut 2012)***	Centers for Medicare & Medicaid Services (CMS) public	Hospital care for Medicare insured (= USA citizens of 65+ age) in 5 clinical areas: heart failure, pneumonia, hip/ knee replacements, CABG, acute myocardial infarction	Single provider model: hospitals	30-day mortality; readmission rate; post-ok haemorrhage; post-ok physiologic / metabolic derangement (16,4%)*
Hospital Readmission Reduction Program (HRRP) USA; since 2012 ***(Kahn et al. 2015; Mellor, Daly, and Smith 2016)***	Centres for Medicare & Medicaid Services (CMS) public	Hospital care for Medicare patients with acute myocardial infarction, heart failure and pneumonia	Single provider model: hospitals	30-day hospital readmissions for acute myocardial infarction, heart failure, pneumonia and hospital acquired conditions (100%)
Hudson Health Plan USA; Since 2004 ***(Chien et al. 2012)***	Hudson Health Plan private; non-profit	Primary care for diabetes patients enrolled in Hudson Health Plan	Single provider model: primary care physicians	Hba1C levels; blood pressure; cholesterol levels; microalbumine levels (46,7%)
Maryland Hospital Acquired Condition Program (Maryland HACP) USA; Since 2009 ***(Calikoglu, Murray, and Feeney 2012)***	State of Maryland public	Hospital care of all patients with hospital acquired conditions (HACs)	Single provider model: hospitals	Hospital acquired conditions (100%)
Medicare Shared Savings Program (MSSP) USA since 2012 ***(Eijkenaar and Schut 2015; (CMS) 2016)***	Centres for Medicare & Medicaid Services (CMS) public	All care for patients assigned to particpating healthcare organisations	Integrated care model: all participating providers involved in targeted care	Blood pressure; HbA1C levels; cholesterol levels (18,2%)
Palo Alto Medical Clinic P4P Program (PAMC P4P) USA; Since 2007 (Chung, Palaniappan, Wong, et al. 2010; Chung, Palaniappan, Trujillo, et al. 2010)	Palo Alto Medical Foundation (PAMF) private; non-profit	Primary care of all patients who visit targeted providers	Single provider model: primary care physicians	Blood pressure; HbA1C levels; cholesterol levels (20,0%)

Name, country, period, and references	Healthcare purchaser	Targeted care	Targeted healthcare providers	Outcome indicators and their contribution (in %) to the performance-related payment size:
Pioneer Accountable Care Organizations (Pioneer ACO) USA; since 2012 ***(Eijkenaar and Schut 2015; (CMS) 2016)***	Centres for Medicare & Medicaid Services (CMS) public	All care for all patients assigned to participating healthcare organisations	Integrated care model: all participating providers involved in targeted care	Blood pressure; HbA1C levels; cholesterol levels (18,2%)
Quality and Outcomes Framework (QOF) UK; Since 2004 ***(Campbell, McDonald, and Lester 2008; Doran et al. 2006; Doran, Fullwood, et al. 2008; Doran et al. 2011; (HSCIC) 2014; Eijkenaar and Schut 2015)***	National Health Service (NHS) public, single purchaser	All primary care for NHS insured (= all UK citizens)	Single provider model: primary care physicians	Blood pressure, HbA1C levels; cholesterol levels; lithium levels (20,8%)
Value Based Purchasing (VBP) USA; since 2012 ***(Eijkenaar and Schut 2015; (CMS) 2017)***	Centres for Medicare & Medicaid Services (CMS) public	Hospital care for CMS insured (= USA low income citizens or 65+ age).	Single provider model: hospitals	30-day mortality, catheter associated urinary tract infections, central line associated blood stream infections, surgical site infections, MRSA or C. Difficile infections and elective deliveries. (2013: 0%; 2014: 25%; 2015: 30%; 2016: 50%; 2017: 50%)
Value Incentive Program (VIP) Korea; since 2007 ***(Kim et al. 2012; Eijkenaar, van de Ven, and Schut 2012; Yang et al. 2016)***	National Health Insurance of Korea (NHIK) public, single purchaser	Hospital care of NHIK insured (= all Korean citizens) in 3 clinical areas: Acute Myocardial Infarction (AMI), Caesar Sections, and acute stroke (since 2012)	Single provider model: hospitals	30-day mortality (30%, AMI only)

TABLE 4.2 | Design features of identified outcome-based payment models

	Indicators				Measurement				Payments					Ref.
	Type of indicators used[A]	No. of indicators (of which outcome indicators)[B]	Extra weight to outcome indicators[C]	Net contribution of outcome indicators to quality score[D]	Scores reported by	Risk-mitigating measures[E]	Publication of scores	Feedback to providers	Incentive types[F]	Requirements for bonus	Requirements for penalty	Requirements for shared savings	Maximum bonus / penalty size[G]	
	Narrow OBPMs													
CQUIN	S, P, O	differs locally	differs locally	differs locally (29% average)	providers	risk-adjustment per indicator	?	?	P	n.a.	differs locally	n.a.	-0,5% (2009) to -2,5% (2012) of contract income	(Kristensen, McDonald, and Sutton 2013; Eijkenaar and Schut 2015)
HQID	S, P, O	AMI: 9 (1) CABG: 8 (3) HF: 4 (0) Pneu: 7 (0) H&K: 6 (3)	no	AMI: 11,1% CABG: 37,5% HF: 0% Pneu: 0% H&K: 50,0%	providers	case mix + exception reporting	yes	yes, annual	B, P	top 20% overall; top 20% improvement	bottom 20%	n.a.	B: +2% on DRG P: -2% on DRG	(Bhattacharyya, Mehta, and Freiberg 2008; Epstein, Jha, and Orav 2014; Eijkenaar and Schut 2015; Eijkenaar, van de Ven, and Schut 2012)
HRRP	O	3(3)	n.a.	100%	?	adjusted for age, sex, co-morbidities	?	?	P	n.a.	below 3-year average readmission rate	n.a.	2012-14: max -1% p/DRG 2015+: max -3% p/DRG	(Kahn et al. 2015; Mellor, Daly, and Smith 2016)
Hudson Health Plan	P, Ox	Diab: 14 (4)	$140/$300 per patient	46,7%	providers	?	no	yes, annual	B	none: fixed price per indicator per patient	n.a.	n.a.	$300,- per patient	(Chien et al. 2012)
Maryland HACP	O	HACs: 49 (49)	no	100%:	providers	corrected for nr of HACs in Y-1	?	?	B, P	?	?	n.a.	B: ? P: -2% of total revenue	(Calikoglu, Murray, and Feeney 2012)
PAMC P4P	S, P, O	15 (3)	no	20,0%	health records	case mix	yes	yes, quarterly	B	achieving minimal target per indicator	n.a.	n.a.	$5000,- per year	(Chung, Palaniappan, Wong, et al. 2010; Chung, Palaniappan, Trujillo, et al. 2010)
QOF	S, P, O	'04: 146 (10) '06: 135 (?) '14: 81 (17)	yes	'14: 20,8%	providers	exception reporting	yes	yes, annual	B	achieving minimal target per indicator	n.a.	n.a.	+25% of budget (after 2014: +17%)	(Campbell, McDonald, and Lester 2008; Doran et al. 2006; Doran, Fullwood, et al. 2008; Doran et al. 2011; (HSCIC) 2014; Eijkenaar and Schut 2015)
VBP	S, P, O	2013: 12 (0) 2014-2015: 15 (3) 2016+: 17 (5)	no	2013: 0% 2014: 25% 2015: 30% 2016: 50% 2017: 50%	providers	corrected for age, sex, CD	yes	yes	B, P	none: general +1% (2013)/ +2% (2017) per DRG	none: general -1% (2013)/ -2% (2017) per DRG	n.a.	B: +1% (2013)/2% (2017) P: - 1% (2013)/-2% (2017) per DRG	(Eijkenaar and Schut 2015; (CMS) 2017)
VIP	P,O	AMI: 6 (1) CS: 1 (0) stroke: 11 (0)	AMI: 1,8x	AMI: 30% CS: 0% Stroke: 0%	claims data	corrected for age	yes	yes, annual	B, P	top 20% overall; top 20% improvement	below threshold (= below 80% best score in Y-2)	n.a.	first phase: B: +1% on DRG P: -1% on DRG second phase B: +1% on DRG P: -1% on DRG	(Kim et al. 2012; Eijkenaar, van de Ven, and Schut 2012; Yang et al. 2016)
	Broad OBPMs													
AQC	S, P, O	pc: 32 (5) sc: 33 (5)	pc: 3x sc: 3x	pc: 35,7% sc: 34,9%	providers	corrected for age, CD	?	yes, monthly	B, SS	>median score; s-shaped relation	n.a.	none	B: +10% of global budget for highest target SS: no max	(Song et al. 2011; Song et al. 2012; Eijkenaar and Schut 2015; Eijkenaar, van de Ven, and Schut 2012)

	Indicators				Measurement				Payments					Ref.
	Type of indicators used[A]	No. of indicators (of which outcome indicators)[B]	Extra weight to outcome indicators[C]	Net contribution of outcome indicators to quality score[D]	Scores reported by	Risk-mitigating measures[E]	Publication of scores	Feedback to providers	Incentive types[F]	Requirements for bonus	Requirements for penalty	Requirements for shared savings	Maximum bonus / penalty size[G]	
	Broad OBPMs													
MSSP	P,O	33 (6)	no	18,2%	patients, providers	no downside risk (option); population correction	?	?	SS	n.a.	n.a.	target on quality indicators	B: 60% of savings (50% if no downside risk) to 7.5% Medicare spending P: 10% of loss	(Eijkenaar and Schut 2015; (CMS) 2016)
Pioneer ACO	P, O	33 (6)	no	18,2%	patients, providers	lim downside risk; popul. correction	?	?	B, SS	?	n.a.	target on quality indicators	B: ? P: ?	(Eijkenaar and Schut 2015; (CMS) 2016)

n.a. = not applicable; ? = unknown

O = outcomes; P = process; S = structure

AMI = acute myocardial infarction; CABG = coronary artery bypass graft; CS = Caesar section; Diab = diabetes; H&K = hip and knee replacement; HACs = hospital acquired conditions; HF = heart failure; pc = primary care; Pneu = pneumonia; sc = secondary care;

AMI = acute myocardial infarction; pc = primary care; sc = secondary care;

AMI = acute myocardial infarction; CABG = coronary artery bypass graft; CS = Caesar section; H&K = hip and knee replacement; HF = heart failure; pc = primary care; Pneu = pneumonia; sc = secondary care;

CD = chronic diseases; HACs = hospital acquired conditions; lim = limited

Effects of OBPMs

Most articles (58) describe effects on quality of care only, 9 articles on healthcare utilization or healthcare costs and 21 articles on both quality and utilization/costs. The follow-up period varies from 9 months to 7 years. Table 4.3 summarizes the effects of OBPMs on quality of care and healthcare utilization/costs.

TABLE 4.3 | Effects of OBPMs on quality of care and healthcare utilization/costs

Model	Quality of care	Healthcare utilization/costs	Number of studies	Downs & Black score: mean (SD)
Narrow OBPMs				
CQUIN	+	?	3	9,0 (1,0)
HQID	mixed*	-	13	11,4 (1,6)
HRRP	+	?	2	9,0 (1,0)
Hudson Health Plan	Mixed	-	2	13,0 (0)
Maryland HACP	+	?	1	10,0 (0)
PAMC P4P	-	?	2	10,5 (2,5)
QOF	+	-**	43	11,9 (1,9)
VBP	-	?	9	11,5 (2,3)
VIP	+	?	3	12,0 (2,0)
Broad OBPMs				
AQC	+	+	10	12,4 (1,1)
MSSP	+	+	2	11,0 (0)
Pioneer ACO	+	+	2	11,0 (0)

Effects are regarded positive when at least 65% of the articles find that a significant improvement in quality of care or reduced health care costs. When the majority of studies found that the quality of care did not improve (or worsened) or healthcare costs increased, we considered the effect negative.

? = unknown

* *After 3 years, the HQID adopted some design changes. In the first phase quality of care improved, the second phase was less successful.*

** *One of the aims of this programme was to increase the income of general practitioners substantially.*

Effects on quality of care

Regarding the effects of the models on quality of care, evidence is available for all 12 models. Of the 88 included studies, 79 targeted quality of care.

Incentivised indicators

All three broad OBPMs showed improvements on the incentivised indicators. Process indicators improved in multiple studies (Song et al. 2011; Song et al. 2012; Song et al.

2014; Eijkenaar and Schut 2015; Barry et al. 2015; Chien et al. 2014), while improvement of outcome indicators was only found for diabetes and vascular care in one study (AQC) (Barry et al. 2015). No improvement was found in outcome indicators for substance use disorder patients (Stuart et al. 2016), emergency department use (both AQC) (Sharp et al. 2013) or hospital readmissions (Pioneer ACO) (McCarthy 2015).

For the narrow OBPMs, five out of nine models showed positive results on the incentivised indicators (CQUIN, HRRP, Maryland HACP, QOF, VIP) (Shlebak et al. 2016; Mellor, Daly, and Smith 2016; Doran et al. 2006; Fleetcroft et al. 2012; Millett, Gray, Saxena, Netuveli, Khunti, et al. 2007; Strong, South, and Carlisle 2009; Kim et al. 2012; Lee et al. 2012; Yang et al. 2016; Doran et al. 2011; Calikoglu, Murray, and Feeney 2012), one showed mixed results (Hudson health plan) (Chien et al. 2012; Chien, Li, and Rosenthal 2010) and in two models no significant effect was found (PAMC, VBP) (Chung, Palaniappan, Trujillo, et al. 2010; Gilman et al. 2015; Figueroa et al. 2016; Ryan et al. 2015; Chee et al. 2016). In the remaining model (HQID), some improvements were observed in the first phase of the programme (first three years), but after some alterations in the design these improvements did not last (Eijkenaar and Schut 2015; Mehrotra et al. 2009; Werner et al. 2011; Epstein, Jha, and Orav 2014; Jha et al. 2012; Ryan 2009).

As in the broad OBPMs, process indicators showed larger improvements than outcome indicators. Five out of nine programmes (CQUIN, HQID, Hudson Health plan, QOF, VIP) reported improvements in certain process indicators (Shlebak et al. 2016; Eijkenaar and Schut 2015; Mehrotra et al. 2009; Werner et al. 2011; Chien, Li, and Rosenthal 2010; Doran et al. 2006; Fleetcroft et al. 2012; Millett, Gray, Saxena, Netuveli, Khunti, et al. 2007; Strong, South, and Carlisle 2009; Campbell et al. 2009; Lee et al. 2011; Vaghela et al. 2009; Millett, Saxena, et al. 2009; Kim et al. 2012; Lee et al. 2012; Yang et al. 2016; Doran et al. 2011), while four (HRRP, Maryland HACP, QOF, VIP) showed improvements in outcomes (Mellor, Daly, and Smith 2016; Alshamsan et al. 2012; Millett, Gray, Saxena, Netuveli, Khunti, et al. 2007; Millett, Saxena, et al. 2009; Vaghela et al. 2009; Yang et al. 2016; Harrison et al. 2014; Kasteridis et al. 2016; Calikoglu, Murray, and Feeney 2012). Two of these could not show improvements in process indicators because these models only included outcome indicators (HRRP and Maryland HACP). Outcome indicators that showed improvements were hospital readmissions after acute myocardial infarction (HRRP) (Mellor, Daly, and Smith 2016), hospital acquired conditions (Maryland HACP) (Calikoglu, Murray, and Feeney 2012), blood pressure and lab results for diabetes and renal disease (both QOF) (Alshamsan et al. 2012; Millett, Gray, Saxena, Netuveli, Khunti, et al. 2007; Millett, Saxena, et al. 2009; Vaghela et al. 2009), mortality after stroke (VIP) (Yang et al. 2016), emergency hospital admissions (QOF) (Harrison et al. 2014) and homecare placements for patients with dementia (QOF) (Kasteridis et al. 2016). However, most outcome indicators did not significantly

improve (Chien et al. 2012; Figueroa et al. 2016; Ryan et al. 2015; Serumaga et al. 2011; Simpson et al. 2011), the mortality rate in particular remaining unaffected (in HQID, QOF, and VBP) (Epstein, Jha, and Orav 2014; Jha et al. 2012; Mehrotra et al. 2009; Ryan 2009; Figueroa et al. 2016; Ryan et al. 2015; Ryan et al. 2016).

While the effects of broad OBPMs on quality of care increased over time (Song et al. 2011; Song et al. 2012; Song et al. 2014; Eijkenaar and Schut 2015), positive effects of narrow OBPMs tended to be short-lived. In two broad OBPMs (AQC and Pioneer ACO), effects on the incentivised indicators increased over the years (Song et al. 2011; Song et al. 2012; Song et al. 2014; Eijkenaar and Schut 2015). In contrast, two narrow OBPMs (HQID and QOF) showed ceiling effects. For HQID this occurred after a significant revision of the incentive structure (Eijkenaar and Schut 2015; Ryan et al. 2012; Shih and Dimick 2013; Werner et al. 2011), while for QOF diabetes and asthma indicators already reached a ceiling after the first year (Campbell et al. 2009). For most of the other indicators in the QOF, ceiling effects emerged after year two or three (Doran et al. 2011; Lee et al. 2011), when many GP practices exceeded the quality thresholds for maximum incentive payments (Fleetcroft et al. 2012). However, the percentage of hospital emergency admissions continued to decrease as a result of the QOF (Harrison et al. 2014).

Relevant provider and patient characteristics

Private providers and providers with low baseline quality scores improved their performance the most (Hudson Health plan, MSSP, Pioneer ACO, QOF, VBP, VIP) (Chien, Li, and Rosenthal 2010; Nattinger et al. 2016; Greene, Hibbard, and Overton 2015; Vaghela et al. 2009; Chatfield 2016; Zhao et al. 2015; Kim et al. 2012; Yang et al. 2016), although some studies concerning the VBP report relatively poor performance of initially low-scoring providers, and in HQID safety net hospitals performed relatively poorly (Eijkenaar and Schut 2015; Ryan et al. 2012; Shih and Dimick 2013; Jha et al. 2012; Figueroa et al. 2016; Gilman et al. 2015; Ryan et al. 2015). Among the narrow OBPMs, three models (HQID, Hudson Health plan, QOF) show that large providers outperform smaller ones (Bhattacharyya, Mehta, and Freiberg 2008; Chien, Li, and Rosenthal 2010; Wang et al. 2006). In the VBP model, this scale effect is mixed (Chatfield 2016; Ramirez et al. 2016; Zhao et al. 2015).

It remains unclear if high-need patients benefit more from OBPMs than other patients. In the AQC, children with special needs benefitted more than others from preventive paediatric care (Chien et al. 2014). In the QOF, quality of care for diabetics with co-morbidities improved more than for those without co-morbidities (Millett, Bottle, et al. 2009). In contrast, mental health centres (AQC), nursing homes (QOF) and hospitals with more Medicare and Medicaid patients (VBP) showed significantly lower quality scores after introduction of a OBPM (Barry et al. 2015; Stuart et al. 2016; Shah et al. 2011;

Zhao et al. 2015). In the Hudson Health Plan, there was no change in quality of care for patients both with and without co-morbidities (Chien et al. 2012).

Effects on healthcare utilization and costs
Regarding the effects on healthcare utilization and healthcare costs, three (out of nine) narrow OBPMs are included (13 studies) in the analysis. Of the broad OBPMs, all three models were included (17 studies).

Healthcare utilization
For five models (AQC, HQID, Hudson Health Plan, Pioneer ACO and QOF), data were available about effects on healthcare utilization. Two out of three narrow OBPMs showed an increase in healthcare utilization. Prescription of preventive drugs increased (antibiotics in HQID (Mehrotra et al. 2009) and antihypertensive drugs in the QOF (Karunaratne et al. 2013)). Moreover, the number of newly diagnosed diabetics who started with medication increased (QOF) (Gallagher et al. 2014). In the Hudson Health Plan, no significant change in healthcare utilization was found (Chien et al. 2012).

Contrary to the narrow OBPMs, the two broad OBPMs showed a reduction in healthcare utilization. For the AQC, reductions among Medicare patients were reported in emergency department use, the use of outpatient care, office visits, minor procedures, imaging and diagnostic tests (McWilliams, Landon, and Chernew 2013). This is in line with the reduction of healthcare utilization found four years after the introduction of the AQC (Song et al. 2014). However, there was no significant impact on the use of pharmaceuticals (Afendulis et al. 2014), while small increases were reported for the use of mental health services (Barry et al. 2015) and emergency departments (Sharp et al. 2013). For the Pioneer ACO programme, a reduction in inpatient services was found (McCarthy 2015).

Healthcare costs
All three broad OBPMs (AQC, MSSP, Pioneer ACO) showed a cost saving based on the incentives of the programme (McWilliams, Landon, and Chernew 2013; Song et al. 2011; Song et al. 2012; Song et al. 2014; Eijkenaar and Schut 2015; McCarthy 2015). The MSSP led to a cost saving of about $385 million within one year, while the Pioneer ACO reached a comparable cost reduction after two years (Eijkenaar and Schut 2015; McCarthy 2015). For the third model (AQC), two out of six studies did not find an effect on healthcare costs (Chien et al. 2014; Sharp et al. 2013), while four studies that were performed later found savings of 1.9%, 3.3%, and 6.8% after 1, 2, and 4 years after introduction, respectively (McWilliams, Landon, and Chernew 2013; Song et al. 2011; Song et al. 2012; Song et al. 2014).

In broad OBPMs, the cost containment effects increased over time. Several studies reported no or small cost reductions in the first years of the AQC programme (Chien et al. 2016; Sharp et al. 2013; Song et al. 2011), while these reductions increased after one or two years (McWilliams, Landon, and Chernew 2013; Song et al. 2012; Song et al. 2014). For the Pioneer ACO programme, one study found similar effects (Eijkenaar and Schut 2015), but another study reported the opposite (McCarthy 2015). For the narrow OBPMs, no longitudinal evaluation studies were available with respect to the impact on costs.

Of the narrow OBPMs, costs increased in all three models for which results are available. This is due to the bonus payments (Ryan 2009; Kruse et al. 2012; Chien, Li, and Rosenthal 2010; Doran, Fullwood, et al. 2008b; Ryan et al. 2016). The HQID does not report any significant effect on healthcare costs, but in the calculation the $17 million that was spent on bonus payments was not taken into account (Ryan 2009; Kruse et al. 2012). Hudson Health Plan, a relatively small programme, spent over $1 million on bonus payments (Chien, Li, and Rosenthal 2010). In the QOF (where a substantial income increase for general practitioners was one of the objectives), over £5 billion was spent in the first seven years of the programme (Doran, Fullwood, et al. 2008b; Ryan et al. 2016), resulting in a 26-40% increase of income for general practitioners (Campbell et al. 2009; Whalley, Gravelle, and Sibbald 2008).

Unintended consequences
For four models (AQC, HQID, Maryland HACP, and QOF), studies were available about effects on non incentivised indicators. For broad OBPMs, data are only available for the AQC. The included studies for this model showed no obvious effect (positive nor negative) on non-incentivised indicators (Chien et al. 2014; McWilliams, Landon, and Chernew 2013). In contrast, for the narrow OBPMs some signs of negative effects exist: while HQID shows no effects on not included indicators (Mehrotra et al. 2009; Ryan et al. 2012), in the Maryland HACP the incidence of non incentivised hospital acquired conditions increased (Calikoglu, Murray, and Feeney 2012). In the QOF there was no change in mortality for either incentivised or non incentivised diseases (Ryan et al. 2016), but (non-incentivised) continuity of care decreased (Campbell et al. 2009). Another study regarding the QOF showed an initial improvement in non incentivised indicators for asthma, diabetes, and vascular diseases, but after two years these effects decreased to below baseline level (Doran et al. 2011).

In three narrow models (HQID, Hudson Health plan, QOF), the effects on ethnic and social disparities were analysed, finding little to no improvement, and sometimes a deterioration. In HQID, the existing gap on process quality closed between blacks and whites, but differences in mortality remained (Epstein, Jha, and Orav 2014). In the Hudson Health Plan, existing disparities in immunisation rates remained (Chien, Li,

and Rosenthal 2010). For QOF, seven out of nine studies found no effects on existing social or ethnic disparities (Doran et al. 2006; Millett, Gray, Saxena, Netuveli, Khunti, et al. 2007; Lee et al. 2011; Alshamsan et al. 2012; Crawley et al. 2009; Millett, Gray, Saxena, Netuveli, and Majeed 2007; Millett et al. 2008). One study showed a decrease between deprived and not deprived patients (Doran, Fullwood, et al. 2008b), while another noticed an increasing gap between socio-economic groups (Simpson et al. 2011).

For the HQID, the HRRP and the QOF (all narrow OBPMs), several studies examine whether or not providers have been trying to abuse the model by directly or indirectly manipulating the performance scores (gaming). In general, there is little evidence that this occurred on a large scale. For HQID and HRRP, no evidence was found that hospitals delay readmissions, alter discharge statuses, limit the access for high-risk patients, or focus on the most profitable measures (Mellor, Daly, and Smith 2016; Nicholas, Dimick, and Iwashyna 2011; Epstein et al. 2014). In the QOF, the generally low levels of exception reporting suggest that large-scale gaming is uncommon (Doran et al. 2006; Serumaga et al. 2011; Doran, Fullwood, Reeves, et al. 2008; Doran et al. 2012; Gravelle, Sutton, and Ma 2010; McDonald and Roland 2009), although some suspect variations in performance scores were noticed (Doran, Fullwood, Reeves, et al. 2008; Doran et al. 2012).

4.4 DISCUSSION

Summary of principal findings

This review provides an evidence synthesis of the characteristics and effectiveness of twelve OBPMs. Based on differences in design features, two groups of OBPMs were distinguised: narrow OBPMs, which only contain explicit financial incentives for objectively measured quality performance; and broad OBPMs, which combine global budgets and risk sharing for multidisciplinary provider groups with explicit financial incentives for quality. Although only three broad OBPMs could be included in this review, their effects on both quality of care and healthcare utilization/costs are particularly favourable when compared to the narrow OBPMs. In addition, these effects improved over time in the broad OBPMs, while the effects of narrow OBPMs tended to be short-lived. We also found that process indicators showed larger improvements than outcome indicators in both groups of OBPMs. Other findings were: larger private providers and providers with initially poor quality scores tended to score better than other providers; high-need patients did not seem to benefit more from OBPMs than other patients; broad OBPMs had little effect on non-incentivised indicators, while there are signs that non-incentivised indicators may deteriorate in the narrow OBPMs; narrow OBPMs did not seem to decrease social or ethnic disparities; and narrow OBPMs do not seem to lead to gaming on a large scale.

Explanations and comparisons to the existing literature

In both groups of OBPMs, process indicators showed larger improvements than outcome indicators. In a way this may be considered disappointing as it raises the question what the value is of focussing financial incentives on outcomes. One explanation is that outcomes are generally more difficult to influence by providers than processes. Another explanation is that improvements in processes may precede improvements in outcomes, especially in the short-term. However, although some studies suggest that the link between processes and outcomes is often not straightforward (Mant 2001). Finally, the improvements on indicator scores could be due to 'signalling power': the implementation of a payment model can lead to increased attention to the incentivised indicators. This attention, rather than the design features of the payment model, could lead to improvements on easy to influence (process) indicators. Nonetheless, the fact that processes improve is positive, given that many earlier evaluations of P4P programmes (which have focused mainly on processes) show mixed effects on process indicators (Eijkenaar et al. 2013).

The broad OBPMs showed increasing improvements on quality indicators over time, while the effects of the narrow OBPMs tend to be short-lived. This may be due to broad OBPMs generally being less prone to ceiling effects due to a design in which explicit incentives based on objectively measured indicators are combined with more general payment mechanisms (i.e. global budgets with risk sharing arrangements). Additionally, the finding that relatively poor performers improve more is another indication of the existence of ceiling effects, which are reported in some of the included models (Eijkenaar and Schut 2015; Ryan et al. 2012; Shih and Dimick 2013; Werner et al. 2011; Campbell et al. 2009; Doran et al. 2011; Lee et al. 2011; Fleetcroft et al. 2012).

We also found that cost savings in broad OBPMs tend to increase over time. In addition, narrow OBPMs typically show increases in healthcare utilization, while broad OBPMs show reductions. These effects might be explained by the additional focus on cost containment in broad OBPMs (i.e. global budgets and risk sharing), while narrow OBPMs focus on quality alone.

Literature on P4P models shows results comparable to our findings on narrow OBPMs: there is evidence that both types of models increase (process) quality of care, although results are mixed and there is no evidence that non-incentivised indicators improve (Eijkenaar et al. 2013). This might be due to similarities in the design: despite the incorporation of more outcome indicators, the working mechanism of narrow OBPMs is often analogous to that of P4P models (i.e. bonuses or penalties for achieving predefined targets with respect to explicitly measured quality indicators).

We found that larger private providers and providers with initially poor quality scores tend to score better than other providers. A possible explanation is that large private providers and providers with low baseline quality have more improvement potential. Moreover, these findings might be influenced by the ceiling effects found in two models (HQID and QOF). In these models, it was relatively easy to achieve a maximum score on some indicators. The distance to these maximum scores from the baseline (i.e. the achieved improvements) is larger in initially low scoring providers. On the other hand, providers with relatively many minority patients or with patients with a lower socioeconomic status are known to have poorer quality metrics. Financial incentives run the risk of exacerbating these disparities across providers. For example: there is evidence that safety net hospitals suffer more from the financial penalties introduced by P4P than other hospitals (Shakir, Armstrong, and Wasfy 2018).

Strengths and limitations

This review has multiple strengths and limitations. The strengths are: 1) this is the most comprehensive review on OBPMs to date, comparing twelve different OBPMs from three different countries; 2) this review has been conducted systematically and multiple data sources were used; and 3) the reviewed studies have a relatively high average level of evidence, since all included quantitative studies adopted a quasi-experimental design. However, as in previous reviews on payment models (Gillam, Siriwardena, and Steel 2012), experimental studies are lacking. This is largely due to the nature of the intervention (i.e. payment models), which often precludes experimental study designs. In addition, for eight of the twelve models, only up to three studies were available. For these models, the results on quality of care or healthcare costs have a limited scientific base.

The use of our definition of OBPMs results in four limitations. First, the required minimum 10% dependency on outcomes set by the definition is an arbitrary cut-off point; it does not take the total size of the performance related reimbursement into account. There is also no evidence for a critical cut-off point in incentive size related to effectiveness. Setting the cut-off point at a lower percentage might have resulted in the inclusion of more programmes, possibly in more countries. However, the 10% threshold seems to allow a reasonably effective distinction between more and less outcome based payment models.

Second, in five of the included programmes (AQC, CQUIN, HQID, VBP, and VIP), we could not determine with absolute certainty if at least 10% of the total incentive payments were always linked to outcome indicators, since these models use separate indicator sets in different geographical regions or care settings. Nevertheless, excluding payment models of which we know that they match our definition in almost all regions or care settings would harm the generalisation of our results. We only

included OBPMs when the information at our disposal consistently confirmed that the model matched our definition and that there were no major differences in specific regions or care settings. This was the case for all five aforementioned OBPMs.

Third, we acknowledge that incentives emanating from payments linked to good scores on outcome indicators might be weaker in included OBPMs with small total incentive payment sizes (e.g. the HRRP) than in excluded models with relatively large total incentive payments but in which less than 10% of these payments are linked to outcomes. However, incorporating the size of these payments into the definition of OBPMs is practically impossible and would lead to an unworkable definition, since the required information is often not available, especially in payment models with complex designs.

A final limitation of our review concerns the generalisation of our findings. First, comparing different outcome measures, used in different OBPMs, is not ideal. Some outcome indicators may have more improvement potential than others, and the existence of clear guidelines can increase this potential. Furthermore, some indicators of the HRRP and the VBP programme overlap, since both programmes are implemented in the context of the USA Medicare programme.

Second, our review includes OBPMs from both in- and outpatient sector, which operate differently. Specifically, they are subject to different payment and billing systems, which affect the incentive structure. In addition, OBPMs in the outpatient sector tend to distribute relatively more money than OBPMs in the inpatient sector. Nonetheless, it is useful to use a broader scope by including both sectors.

Third, the effects of the payment models are likely to be influenced by contextual factors. The introduction of OBPMs is often part of a larger policy package, such as increased registration, public reporting or implementation of feedback systems. Effects can also be influenced by the healthcare system of the involved country. The fact that models are from different countries leads to challenges in drawing conclusions. However, it must be highlighted that nine out of the twelve models in this review are from the USA. Although this makes a comparison between these nine models easier, the USA is a country with exceptionally high healthcare costs. Positive effects on healthcare costs might therefore be easier to achieve than in other countries. Consequently, extrapolation of findings from USA-based studies to other healthcare systems is hard.

Conclusions

OBPMs are at the centre of the debate on the future of healthcare reimbursement. It is one of the theoretical underpinnings of the movement towards value-based-

healthcare which seeks for more quality of care and value against the 'lowest' possible costs (Porter and Teisberg). We conclude that an increased focus on outcome indicators alone is unlikely to result in an increased effectiveness of payment models: other design features also influence the effects on quality of care and healthcare costs. Specifically, our main findings suggest that OBPMs which combine global payments and risk-sharing with explicit bonuses or penalties based on (outcome) indicator scores have most potential to contribute to value. Based on our results, these 'broad' OBPMs seem to be more (cost-)effective than the 'narrow' OBPMs, as in the latter group evidence of improved quality is less consistent and tends to be short lived, and evidence for decreases in healthcare costs is lacking. Despite the limitations of our approach and the fact that we still know little about the interaction between costs and quality, we feel that we can recommend broad OBPMs. However, given that we could only include three broad OBPMs, which have all been implemented more recently than the 'narrow' OBPMs and all in the USA, more rigorous evaluations of broad OBPMs are required to strengthen this conclusion, preferably in a different context than that of the USA.

Appendices | Supplementary material

APPENDIX 4.1 SEARCH STRING

(Outcome[it] OR quality*[it] OR performance*[it] OR value*[it] OR readmission*[it] OR mortality [it] OR complication*[it])*

AND

(incentiv[it] OR pay*[it] OR fund[it] OR funding[it] OR funds[it] OR remunerat*[it] OR reimburs*[it] OR financ*[it] OR fee[it] OR fees[it] OR purchas*[it] OR buy*[it] OR contract[it] OR contracts[it] OR contracting[it])*

AND

(model[tiab] OR models[tiab] system[tiab] OR systems[tiab] scheme[tiab] OR schedule[tiab] OR reform[tiab] OR reforms[tiab] OR program[tiab] OR programme[tiab] OR programs[tiab] OR programmes[tiab]OR framework*[tiab] OR contract[tiab] OR contracts[tiab] OR contracting[tiab] OR project*[tiab])*

APPENDIX 4.2 | Consulted agencies

Agency name	*Country*
Agency for Healthcare Research and Quality (AHRQ)	USA
Australian Institute of Health and Welfare (AIHW)	Australia
Bertelsmann Foundation	Germany
Commonwealth Fund	USA
Health Foundation	UK
Institute for Healthcare Improvement (IHI)	USA
Institute for research and information in health economics (IRDES)	France
King's Fund	UK
Leapfrog Group	USA
National institute for Health and Care Excellence, (NICE)	UK
National institute for Health and Medical research (INSERM)	France
Nuffield Trust	UK
Organisation for Economic Co-operation and Development (OECD)	-
Robert Bosch Foundation	Germany
Robert Wood Johnson Foundation (RWJF)	USA
Swedish Institute: College of Health Sciences	Sweden
United States Health Information Knowledgebase (USHIK)	USA
World Health Organisation (WHO)	-

APPENDIX 4.3 | Consulted experts

Prof. M. Rosenthal	Professor of Health Economics and Policy and Associate Dean for Diversity at Harvard T.H. Chan School of Public Health, Cambridge, Massachusetts, USA
Prof. F.E. Schut	Professor of Health Economics and Health Policy at the Erasmus School of Health Policy and Management (iBMG), Erasmus University of Rotterdam, The Netherlands
Dr. F. Eijkenaar	Assistant Professor at the Erasmus School of Health Policy and Management (iBMG), Erasmus University of Rotterdam, The Netherlands

APPENDIX 4.4 | Quality assessment score

Quantitative studies (Downs & Black score)	Total score	Scores on individual items				
		Reporting	External validity	Internal validity		Power
				Bias	Confounding	
	(max 32)	*(max 11)*	*(max 3)*	*(max 7)*	*(max 6)*	*(max 5)*
Afendulis 2014	14	7	1	4	2	0
Alshamsan 2012	14	7	0	4	3	0
Barry 2016	13	7	1	3	2	0
Bhattacharyya 2008	10	5	0	4	1	0
Calikoglu 2012	10	4	0	3	3	0
Campbell 2009	13	6	0	4	3	0
Chatfield 2016	9	4	1	3	1	0
Chien 2010	13	7	0	3	3	0
Chien 2012	13	7	0	3	3	0
Chien 2014	11	5	1	3	2	0
Chung 2010a	13	4	2	4	3	0
Chung 2010b	8	4	0	3	1	0
Crawley 2009	10	6	0	3	1	0
Dalton 2011	15	7	1	4	3	0
Das 2016	12	6	2	3	1	0
Doran 2006	14	8	1	4	1	0
Doran 2008a	13	7	1	4	1	0
Doran 2008b	11	6	1	4	0	0
Doran 2011	15	7	1	4	3	0
Doran 2012	12	5	2	2	3	0
Epstein 2014a	14	8	0	3	3	0
Epstein 2014b	12	7	0	3	2	0
Figueroa 2016	17	9	3	3	2	0
Fleetcroft 2012	12	7	0	4	1	0
Gallagher 2014	15	8	2	4	1	0
Gemmell 2009	16	9	1	4	2	0
Gilman 2015	11	5	1	3	2	0
Gravelle 2010	13	8	1	3	1	0
Greene 2015	11	7	0	3	1	0
Guthrie 2006	10	4	1	4	1	0
Harrison 2014	12	7	1	3	1	0
Jha 2012	12	7	0	3	2	0

Quantitative studies (Downs & Black score)	Total score	Scores on individual items				
		Reporting	External validity	Internal validity		Power
				Bias	Confounding	
	(max 32)	*(max 11)*	*(max 3)*	*(max 7)*	*(max 6)*	*(max 5)*
Kahn 2015	10	3	0	4	3	0
Karunaratne 2013	10	6	0	3	1	0
Kasteridis 2016	14	8	1	3	2	0
Kendrick 2015	10	6	0	3	1	0
Kontopantelis 2012	10	5	0	3	2	0
Kontopantelis 2016	10	5	1	3	1	0
Kristensen 2013	10	6	0	2	2	0
Kruse 2012	14	7	2	3	2	0
Lee 2011	9	5	0	3	1	0
Lee 2012	14	6	1	4	3	0
MacBride-Stewart 2008	11	6	1	3	1	0
McWilliams 2013	13	8	1	3	1	0
Mellor 2016	8	4	0	3	1	0
Millett 2007a	11	7	0	3	1	0
Millett 2007b	12	7	0	3	2	0
Millett 2008	12	6	0	3	3	0
Millett 2009a	11	6	0	3	2	0
Millett 2009b	8	4	0	3	1	0
Nattinger 2016	11	5	1	3	2	0
Nicholas 2011	11	5	0	4	2	0
Ramirez 2016	11	6	0	3	2	0
Ryan 2009	12	5	1	4	2	0
Ryan 2012	11	5	1	3	2	0
Ryan 2014	10	4	1	3	2	0
Ryan 2015	12	7	0	3	2	0
Ryan 2016	12	6	1	3	2	0
Serumaga 2011	12	7	1	3	1	0
Shah 2011	13	5	2	3	3	0
Sharp 2013	13	6	1	4	2	0
Shih 2014	10	6	0	3	1	0
Shlebak 2016	8	5	0	2	1	0
Simpson 2011	8	4	0	3	1	0
Song 2011	13	7	1	3	2	0
Song 2012	11	5	1	3	2	0
Song 2014	11	5	1	3	2	0
Strong 2009	13	5	2	4	2	0
Stuart 2016	13	6	1	3	3	0
Vaghela 2009	12	7	1	2	2	0
Wang 2006	13	6	2	3	2	0
Werner 2011	9	4	0	4	1	0
Whalley 2008	12	6	0	4	2	0
Yang 2016	10	6	0	3	1	0
Zhao 2015	10	5	0	3	2	0

Qualitative studies (CASP method)	scores on individual items									
	1	2	3	4	5	6	7	8	9	10
Campbell 2008	yes	yes	can't tell	can't tell	yes	yes	yes	yes	yes	highly valuable
Chien 2016	no	yes	can't tell	yes	can't tell	yes	yes	can't tell	no	limited
Edwards 2007	yes	yes	yes	yes	yes	can't tell	yes	no	yes	valuable
Hannon 2012	yes	yes	can't tell	yes	no	yes	yes	can't tell	yes	limited
Lester 2013	yes	yes	can't tell	no	no	can't tell	yes	no	yes	valuable
Maisey 2008	yes	yes	can't tell	no	can't tell	yes	no	yes	yes	limited
McDonald 2009	yes	yes	can't tell	no	can't tell	can't tell	yes	no	no	valuable
Norman 2014	yes	yes	can't tell	yes	can't tell	can't tell	yes	no	yes	limited

Reviews (CASP method)	scores on individual items									
	1	2	3	4	5	6	7	8	9	10
Gillam 2012	no	no	yes	yes	can't tell	clearly described	can't tell	yes	yes	no
Mehrotra 2009	yes	can't tell	yes	yes	no	clearly described	can't tell	can't tell	yes	no

APPENDIX 4.5 | **List of identified OBPMs**

Model name	*Country*	*Effect study available?*
Alternative Quality Contract (AQC)	USA	yes
Bridges to Excellence	USA	no
Chipra	USA	no
Commissioning for Quality and Innovation (CQUIN)	UK	yes
Geisinger Health System	USA	no
General Practitioners Consortiums	UK	no
Georgia Blues	USA	no
Health eHearts	USA	no
Home Health P4P	USA	no
Hospital Acquired Conditions Reduction Program (HACRP)	USA	no
Hospital Quality Incentive Demonstration (HQID)	USA	yes
Hospital Readmissions Reduction Plan (HRRP)	USA	yes
Hudson Health Plan	USA	yes
Long Island Health Network P4P	USA	no
Maryland Hospital Acquired Conditions Program	USA	yes
Medicare shared savings program (MSSP)	USA	yes
Minnesota nursing home P4P	USA	no
New Hampshire Accountable Care Organisations	USA	no
Norway P4P model	Norway	no
Oregon Salem	USA	no
Palo Alto Medical Clinic P4P (PAMC P4P)	USA	yes
Pharmacist P4P	USA	no
Pioneer accountable care organisations (Pioneer ACO)	USA	yes
PROMETHEUS payments	USA	no
Quality Incentive Program end-stage renal disease	USA	no
Quality and Outcome Framework	UK	yes
Value Incentive Program (VIP)	Korea	yes
Value-Based Purchasing	USA	yes
Wellmark IVF P4P	USA	no

CHAPTER 5

Density of patient-sharing networks: impact on the value of Parkinson care

F.P. Vlaanderen, Y. de Man, M.A.C. Tanke, M. Munneke, F. Atsma, M.J. Meinders, P.P.T. Jeurissen, B.R. Bloem, J.H. Krijthe, A. S. Groenewoud, 'Density of patient-sharing networks: impact on the value of Parkinson care'. *International Journal of Health Policy and management*; under review

ABSTRACT

Background: Optimal care for Parkinson's disease (PD) requires coordination and collaboration between providers within a complex care network. Individual patients have personalised networks of their own providers, creating a unique informal network of providers who treat ('share') the same patient. These 'patient-sharing networks' differ in density, i.e. the number of identical patients they share. Denser patient-sharing networks might reflect better care provision, since providers who share many patients might have made efforts to improve their mutual care delivery. We evaluated whether the density of these patient-sharing networks affects patient outcomes and costs.

Methods: We analysed medical claims data from all people with PD in the Netherlands between 2012 and 2016. We focused on seven professional disciplines that are commonly involved in Parkinson care. We calculated for each patient the density score: the average number of patients that each patient's providers shared. Density scores could range from 1·00 (which might reflect poor collaboration) to 83·00 (which might reflect better collaboration). This score was also calculated at the hospital level by averaging the scores for all patients belonging to a specific hospital. Using logistic and linear regression analyses we estimated the relationship between density scores and health outcomes, healthcare utilization, and healthcare costs.

Results: The average density score varied considerably (average 6·7, SD 8·2). Adjusted for confounders, higher density scores were associated with a lower risk of PD-related complications (OR: 0·901; p<0·001) and with lower healthcare costs (coefficients: -0·018, p=0·005). Higher density scores were associated with more frequent involvement of neurologists (coefficient 0·068), physiotherapists (coefficient 0·052) and occupational therapists (coefficient 0·048) (p-values all <0·001).

Conclusion: Patient sharing networks showed large variations in density, which appears unwanted as denser networks are associated with better outcomes and lower costs.

5.1 INTRODUCTION

Achieving optimal care for patients with a chronic neurological condition is challenging.(Bloem et al. 2020) Optimal management requires a multidisciplinary approach, a complex array of treatment options and a long follow-up. Networks of healthcare providers have proven to be useful for improving coordination and organization of care.(Willis et al. 2013) Interestingly, healthcare consists of more than such formal professional networks, because individual patients also build their own informal personalised networks: they choose (or are being allocated to) their own set of healthcare providers, leading to a unique network of providers who treat ('share') the same patient.(Landon et al. 2012) These so called 'patient-sharing networks' of healthcare providers will typically differ in 'density', i.e. in the number of identical patients they share.(Pollack et al. 2012)

Denser patient-sharing networks, i.e. networks of providers who share relatively more patients with each other, might result in better care provision. Providers in a dense network might communicate and cooperate better,(Foy et al. 2010; Barnett et al. 2011) or know each other through referrals.(Barnett et al. 2011) This could improve the coordination and organization of care for their patients.(Pollack et al. 2012) Increased patient-sharing within group practices has been positively associated with patient-reported care coordination. (Moen and Bynum 2019) Positive effects of dense networks might be expected especially among patients with chronic conditions, since these patients likely benefit most from integrated, well-organized care delivery.(Bloem et al. 2020)

In this study, we aimed to study the effects of network density in the context of a chronic neurological condition, using Parkinson's disease (PD) as an illustrative example. The care for people with PD is complex, because many different healthcare providers are involved, many of whom work in different echelons of healthcare (primary care, hospitals, long term care).(Bloem et al. 2010; Radder et al. 2018) Most people with PD visit neurologists, physiotherapists, occupational therapists and speech & language therapists. Dieticians and psychologists are also frequently involved, and in advanced PD the number of involved disciplines can be as high as 18.(Bloem et al. 2010) This provides great challenges to the coordination and organization of multidisciplinary care. Organising care delivery in professional networks of specifically trained healthcare providers at a regional level leads to better collaboration and fewer disease complications, (Keus et al. 2014) but there is no evidence that the density of patient-sharing networks improves care delivery and leads to better outcomes. In this study, we therefore assessed the relation between the density of patient-sharing networks and health outcomes, healthcare utilization and healthcare costsfor patients with PD. Specifically, we aimed to investigate (a) to what extent patient-sharing networks in PD vary in density in current daily clinical practice (assuming that large variations are

generally unwanted); and (b) if denser patient-sharing networks are associated with better health outcomes, lower healthcare utilization and lower healthcare costs.

5.2 METHODS

Data

We analysed medical claims data from all people with PD in the Netherlands between 2012 and 2016. These data were made available through Vektis, a not-for-profit organization that collects all claims data for all Dutch healthcare insurance companies. (Vektis 2020) All Dutch inhabitants are obliged by law to have a private healthcare insurance, which is partially paid for by the government. Insurance companies are obliged to accept everybody (against the same price), and the compliance among Dutch citizens to this health insurance obligations is as high as 99·8% (Zorgwijzer 2019). The database of Vektis therefore contains the claims data of 17·4 million people. (CBS 2020) These claims data concern all primary and secondary care, plus the costs for nursing home residency. The Vektis data also include the date when a person died. We successfully used this same Vektis database in a previous analysis where we demonstrated the added value of professional networks of physiotherapists who were specifically trained to treat patients with PD.(Ypinga et al. 2018)

Study sample

We included all 48,769 Dutch insured citizens who had at least one diagnostic related group code (DRG code) of PD since January 2008. This selection was part of the preparation of our database and was performed by Vektis. Data of individual patients were included in the analyses from the moment that the first PD DRG appeared for that patient. The first PD DRG defines the moment of diagnosis by a neurologist. The same approach was used in earlier research on PD care in the Netherlands.(Ypinga et al. 2018; Vlaanderen et al. 2019) The included patients were given a unique random identifier by Vektis. The key to the identifier was not available to the researchers.

Similar to previous research on claims data for PD care,(Vlaanderen et al. 2019) we included the PD-related claims data of neurologists, specialized PD nurses (both included at the hospital level, since some hospitals tend to wrongly claim on just one neurologist or specialised nurse while care is provided by many), physiotherapists, occupational therapists, speech & language therapists, dieticians, and psychologists. These are the healthcare providers that are most frequently involved in PD management.(Bloem et al. 2010; Keus et al. 2014; Kalf et al. 2008; Sturkenboom et al. 2008; van Asseldonk et al. 2012) Every healthcare provider was given a random identifier in a similar way as the patients.

The definition of density

In order to assess how 'dense' a patient-sharing network is, we used the model for care density defined by Pollack et al(Pollack et al. 2012):

$$density\ score = \frac{\sum_{i=1}^{m} w_{p,i}}{n_p(n_p - 1)/2}$$

where n_p is the number of distinct healthcare providers that patient p saw, m is the total number of possible pairs of these healthcare providers, and $w_{p,i}$ is the number of shared patients for each pair of healthcare providers. The numerator is the total number of instances of patient sharing over the study period among a patient's providers. The denominator is the total number of pairs of healthcare providers for that patient. The higher the density score, the more patients the involved providers share. A visual example of this method is given in Figure 5.1. More details of this method can be found in Pollack et al.(Pollack et al. 2012)

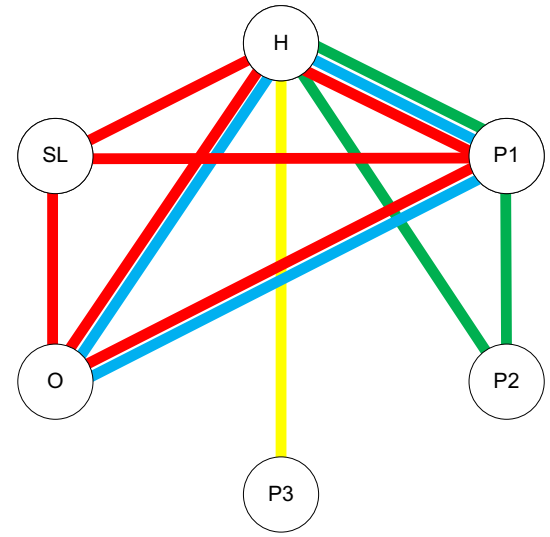

FIGURE 5.1 | Visual example for a situation in which six healthcare providers

(hospital H, speech & language therapist SL, occupational therapist O, and physio-therapists P1, P2 and P3) share four patients (blue, red, yellow and green). The density score of the provider network of the blue patient = (2+2+3)/(3(2-1)/2) = 2·33, while density scores are 1·67, 1·67, and 1·00 for the red, green, and yellow patient respectively.*

Comparing density scores

First, the density score per patient was calculated for 36,639 patients. Density scores ranged from 1·00 (which might reflect poor collaboration) to 83·00 (which might reflect better collaboration). For the remaining 12,130 patients it was impossible to calculate a density score because they visited either zero, just one or, due to missing values in the dataset, an unknown number of healthcare providers (Table 5.1).

We then calculated the average density score per hospital across their entire PD patient population, to see if average density scores varied between hospital populations. We therefore assigned all people with PD to the hospital from which they had received most of their hospital care, i.e., from which they had the most neurologist and specialized PD nurse claims. We were only able to calculate the average scores for 108 out of all 136 Dutch hospitals, since 28 hospitals did not have any people with PD assigned to them. These 28 hospitals were probably hospitals which fused shortly after the start of our time span with other hospitals, or were large specialised medical clinics which do not treat PD.

To visualize the differences in density scores, we selected the three hospital populations with the highest and three other hospital populations with the lowest density score. The average density score per hospital population was positively influenced by the number of assigned patients. Regarding the lowest density scores, we therefore only considered hospital populations with at least 369 patients, which was the size of smallest hospital population with the highest density score. We visualized the networks of the selected hospital populations by plotting the healthcare providers that shared mutual patients with t-distributed Stochastic Neighbour Embedding (t-SNE).(van der Maaten and Hinton 2008)

Outcomes, utilization and costs

Our next main aim was to assess the relation between density scores and health outcomes, healthcare utilization and healthcare costs. For this purpose, we defined the following outcome measures. *Health outcomes* were defined as the occurrence of any one or more of three PD-related complications (i.e. a claimed DRG for pneumonia, orthopaedic injury or hospital admission for PD)(Ypinga et al. 2018; Vlaanderen et al. 2019) and mortality. *Healthcare utilization* was defined as the number of DRGs (neurologist and specialized PD-nurses) or visits (allied healthcare providers) to the included healthcare providers. *Healthcare costs* were defined as the sum of the prices of the claims. We calculated healthcare costs separately with and without the costs of the DRGs of PD-related complications.

Subsequently, we used regression analyses for each outcome. To assess the association between density scores and health outcomes we used logistic regression models since

the dependent variables were all dichotomous; the associations with utilization and costs were performed with linear regression models. In the linear regression analyses we log-transformed the dependent variables since the effects on utilization and costs were multiplicative rather than additive and the residuals were closer to the normal distribution if the dependent variables were log-transformed. For similar reasons, and to unify our results and simplify the interpretation, we log-transformed the density scores in all regression analyses.

In all regression analyses, we adjusted for age, sex, the duration of the disease, the number of healthcare providers per patient, the average number of patients per healthcare provider in the patient's network, and the follow-up time. Age, sex and the duration of the disease might influence the dependent variables, and these were therefore added to the regression model as independent variables. For duration of the disease, we added an extra variable indicating if a patient had PD-related claims in 2008 or not. This was done to cope with the limitation that our dataset does not contain data prior to 2008, which would otherwise result in underestimation of disease duration. The number of healthcare providers per patient and the average number of patients per provider in the patient's network were added in a similar way, since these variables appeared to have a correlation with density of -0.093 and 0.254 respectively (p-values both < 0.001).

For all regression models, we excluded patients with either zero, or one or an unknown number of healthcare providers. In the logistic regression models on PD-related complications, we additionally excluded patients of which we did not have the full follow-up time available (five years), since these patients would have had less time to develop a complication. This resulted in 13,129 included patients. In the linear regression models, we adjusted for follow-up by defining the dependent variables as averages per month. Patients with less than six months follow-up were excluded to avoid outliers. This resulted in 35,414 included patients for the linear regression analyses on healthcare costs. For the linear regression analyses on utilization, only patients could be included that had claims from the designated healthcare providers. We included 33,703 for neurology utilization, 33,474 for physiotherapist utilization, and 14,534 for occupational therapist utilization. For speech and language specialist utilization 8,895 patients could be included, and the numbers for dieticians and psychologist utilization were 6,490 and 6,437 respectively.

Secondary analysis

Our results might be influenced by the activities of the Dutch nationwide ParkinsonNet healthcare network. Covering the entire country, ParkinsonNet is a Dutch not-for-profit organization, supporting regional provider networks of medical and allied healthcare professionals specialized in the management of patients with PD. The ParkinsonNet

approach stimulates concentration of care among the specifically trained professionals (which influences density scores), but also develops guidelines, stimulates collaboration and provides education to healthcare providers.(Keus et al. 2014; Nijkrake et al. 2010) These efforts have led to improved health outcomes, and decreased healthcare costs. (Keus et al. 2014)

To assess the relation between the density score and membership of the ParkinsonNet network, we identified for all included providers if they were a ParkinsonNet member or not. Since the claims of neurologists and specialized PD-nurses in our data set were only available at the hospital level, and because membership of ParkinsonNet is individual, we excluded the neurology DRGs from this analysis. Subsequently, we performed an additional linear regression analysis on the log-transformed density scores to identify if there was a correlation between the log-transformed density score and the percentage of visits to ParkinsonNet healthcare providers. The variables 'number of healthcare providers per patient' and 'the average number of patients per provider in the patient's network' were added as independent variables, since they influence the dependent variable (log-transformed density score). After exclusion of the hospital claims and exclusion of patients with (then) zero, one or an unknown number of healthcare providers, 36,639 patients could be included in this linear regression model.

Ethical Statement

This study was approved by the institutional review board of the Radboud University Medical Centre with a waiver of consent for participants in the study (file number 2019-5106).

Data availability statement

The data that support the findings of this study are available from the corresponding author, upon reasonable request. However, the original claims data belong to Vektis. Permission of Vektis is required before original claims data can be made available due to privacy laws.

5.3 RESULTS

Variation in density

The characteristics of the study sample are presented in Table 5.1. The average density score varied considerably. At the individual patient level, the average score was 6·7 (SD: 8·2). At the level of hospital populations, it was 3·9 (SD: 1·8). This difference in average scores arose because many hospitals had very few patients assigned (17 hospitals had less than 100 patients).

TABLE 5.1 | General characteristics of the study sample (n = 48,769)

Average age (in 2012)		71·7 years (SD: 10·1)
Sex		58·9 % men
Time since diagnosis* (in years, median (IQR))		5·5 (4·5 – 7·7)
Follow-up time (in years, mean (SD))		3·2 (1·7)
Number (%) of patients with complete follow-up		16,404 (33·6)
Number (%) of patients with less than 6 months follow-up		3,544 (7·3)
Number (%) of providers per patient during follow-up:	0 or unknown	2,222 (4·6)
	1	9,908 (20·3)
	>1	36,639 (75,1)
	Median (IQR)	3,0 (2,0 – 5,0)
Number (%) of patients visiting	Hospital (neurologist/specialized nurse)	40,980 (84,0)
	Physiotherapist	34,496 (70,7)
	Occupational therapist	14,944 (30,6)
	Speech & language therapist	9,052 (18,6)
	Dietician	6,600 (13,5)
	Psychologist	7,549 (15,5)
Number of included	Hospitals	136
	Physiotherapists	14,743
	Occupational therapists	1,232
	Speech & language therapists	984
	Dieticians	1,286
	Psychologists	238
PD-related healthcare costs per year per patient		
Costs of complications excluded (Mean (SD))		$ 2004,99 (3083·59)**
Costs of complications included (Mean (SD))		$ 2153,47 (3145,99)**

*SD = Standard Deviation; IQR = inter quartile range; * = calculated to date of death or up to December 31st 2016 (the last day of the dataset); ** = euro-dollar conversion per March 13th 2019*

The t-SNE visualization of the variation between hospital populations is shown in Figure 5.2. For the top three hospital populations, providers shared more patients with the hospital (bigger dots) and also with each other (more clusters, more darker lines).

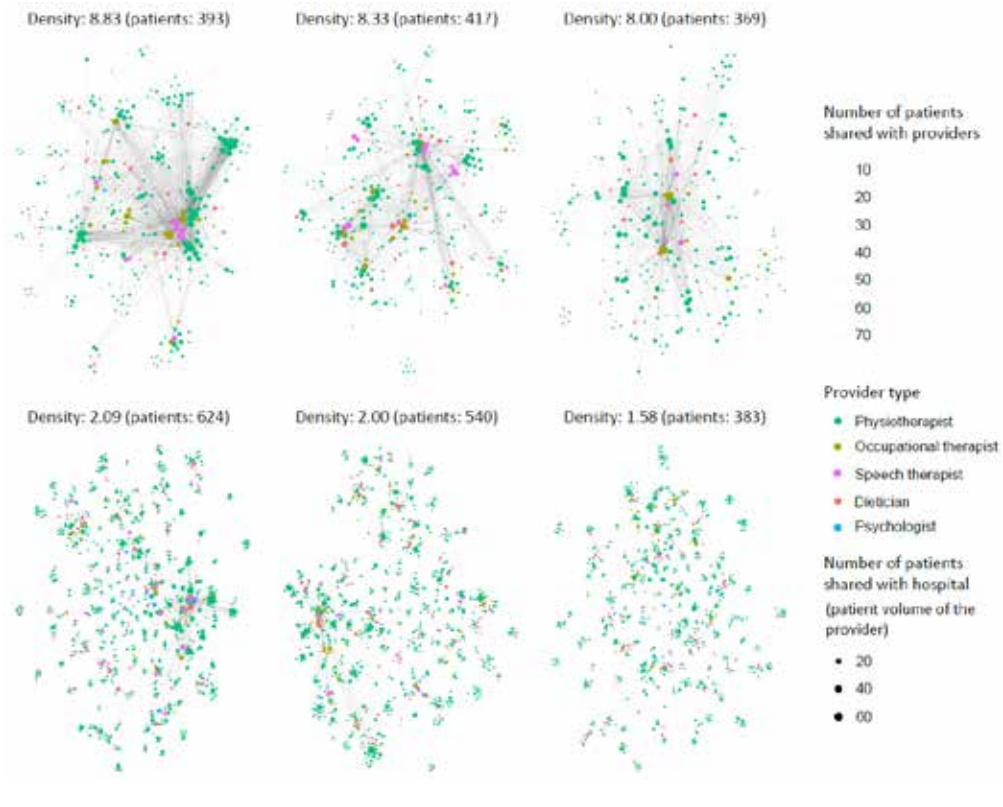

FIGURE 5.2 | **t-SNE visualization of top three and bottom three hospital populations in terms of average density scores**

Each dot represents a provider; the colour represents provider type; the size of the dot represents the number of shared patients with the hospital (the hospital itself is not shown); lines and relative location between the dots represent the number shared patients with each other (only when 10+ patients are shared, a line is shown; when more patients are shared, the line is darker.

Regression models on health outcomes, utilization and costs

Table 5.2 shows that higher density scores were associated with lower incidences of PD-related complications (OR: 0·901, p-value <0·001). A doubling of the density score was associated with lower odds of complications of approximately 1-2^log(0·901) ≈ 7·0%. In a similar way, denser patient-sharing networks were associated with a lower occurrence of pneumonias and orthopaedic injuries, but not with lower PD-related hospitalization.

TABLE 5.2 | **Adjusted estimates relating log(density score) to health outcomes, healthcare utilization and healthcare costs**

Health outcomes	Odds ratios	Confidence interval (95%)	P-value
Incidence of PD-related complications			
Incidence of pneumonia	0·926	0·889 – 0·964	<0·001
Incidence of orthopedic injuries	0·899	0·864 – 0·936	<0·001
Incidence of PD-related hospitalizations	1·023	0·971 – 1·079	0·392
Incidence of all PD-related complications	0·901	0·862 – 0·941	<0·001
Mortality	0·962	0·926 – 1·000	0·052
Healthcare utilization	**Coefficients**	**Confidence interval (95%)**	**P-value**
Log-transformed neurologist utilization	0·068	0·062 – 0·075	<0·001
Log-transformed physiotherapist utilization	0·052	0·038 – 0·065	<0·001
Log-transformed occupational utilization	0·048	0·028 – 0·068	<0·001
Log-transformed speech & language therapist utilization	0·024	-0·009 – 0·057	0·156
Log-transformed dietician utilization	-0·013	-0·043 – 0·017	0·409
Log-transformed psychology utilization	-0·032	-0·061 – -0·003	0·029

All values are adjusted for the effects of age, sex, the duration of the disease, the number of healthcare providers per patient, the average number of patients per healthcare provider in the patient's network and the differences in follow-up time.

Higher density scores were associated with more frequent involvement of neurologists, physiotherapists and occupational therapists (coefficients of 0·068, 0·052 and 0·048 respectively, p-values all <0·001), but to less frequent involvement of psychologists (coefficient of -0·032; p-value 0·029). A doubling of the density score was associated with an increase 4·8% for neurologist utilization, 3·7% for physiotherapist utilization, and 3·4% for occupational therapist utilization. Similarly, an exponential increase in density score was associated with decreased psychologist utilization of approximately 2·2%. For speech & language therapists and dieticians we found no associations.

Healthcare costs seemed to be negatively associated with density scores (Table 5.2). A doubling of the density score was associated with reduced healthcare costs of approximately 1·2% (complications excluded) to 2·1% (complications included). Compared to the average healthcare costs for PD (Table 5.1), this would equate to an annual reduction of $24,06 to $45,22 per patient (over 36,639 patients, this corresponds

with $0·9 million to $1·7 million). The appendices show the complete regression analyses on health outcomes (A), healthcare utilization (B), and healthcare costs (C).

Secondary analysis

A high percentage of visits to ParkinsonNet members was associated with higher density scores (coefficient of 1·164; p-value <0·001). Patients who exclusively consulted ParkinsonNet professionals had an approximately exp(1,164) ≈ 3·2 times higher density score compared to patients who never consulted ParkinsonNet members. The linear regression analysis is included as appendix 5.3.

5.4 DISCUSSION

Our aim was to identify whether provider networks for individual people with PD differ in density and, if so, whether denser patient-sharing networks would be associated with higher value of care. These questions were addressed in a unique cohort of all people with PD in the Netherlands followed over a 5-year timeframe. Several findings emerged. First, we identified substantial differences in the density scores between patient-sharing networks. These differences were found both at the level of the individual patient and at the level of hospital populations. Second, our analyses show that denser patient-sharing networks are associated with a lower occurrence of PD-related complications, especially fewer pneumonias and orthopaedic injuries. Third, denser patient-sharing networks are associated with more common involvement of neurologists, physiotherapists and occupational therapist services, but also with somewhat lower utilization of psychologists. Fourth, denser patient sharing networks are associated with a small decrease in healthcare costs for PD management. Finally, our secondary analysis shows a strong correlation between the density score and the percentage of visits to providers associated with ParkinsonNet, a Dutch network of specialised healthcare professionals. This suggests that the observed effects might be influenced by the efforts of this integrated network approach.

Comparison of our study to previous studies is difficult, since no prior work assessed patient-sharing network densities nor their effects in a PD context. Two large studies about the density of patient-sharing networks in the US showed no impact on quality of care, while healthcare utilization and costs increased.(Barnett et al. 2012; Landon et al. 2018) Both studies focused on the densities of general Medicare insured patients, rather than on patients with chronic conditions for whom the bests results can be expected. When focusing on patients with chronic conditions, the average density values that we identified at the individual patient level are comparable with density values of patient-sharing networks of patients with diabetes, congestive heart failure, COPD and cancer.(Pollack et al. 2012; Pollack et al. 2014; Pollack et al. 2015) This supports the validity of our approach. Research on these other chronic conditions identified

similar associations between care density and health outcomes: cancer survivors with denser networks are hospitalized less often,(Pollack et al. 2014) while diabetes patients with denser networks have a lower risk of being readmitted to hospital and to experience potentially avoidable complications.(Pollack et al. 2015) The last study found no positive associations in the context of congestive health failure and COPD, and even found negative associations with some other quality measures. Additionally, denser patient-sharing networks have been linked to reduced healthcare costs for patients with congestive heart failure,(Pollack et al. 2012) diabetes,(Pollack et al. 2012) and cancer.(Pollack et al. 2014)

Our method has several strengths. First, we followed a clear and predefined set of analyses. Second, our dataset contained all diagnosed patients in the country, which limits the risk of selection bias. However, some selection bias might have been introduced because inclusion was based on the PD DRG. Our sample may have included cases for whom the initial diagnosis of PD turned out to be incorrect. PD can be difficult to diagnose in early disease stages, with reported diagnostic error rates of >10%,(Skinner, Scott, and Martin 2016) e.g. because forms of atypical parkinsonism can present like PD.(Hijdra, Koudstaal, and Roos 2012) Because our sample matches well with the population characteristics of other studies in terms of prevalence,(Hijdra, Koudstaal, and Roos 2012) age,(Ypinga et al. 2018; Hijdra, Koudstaal, and Roos 2012) division of sex,(Ypinga et al. 2018; Hijdra, Koudstaal, and Roos 2012; Jankovic, Hurtig, and Eichler 2019) and healthcare use,(Ypinga et al. 2018) we do not think this has greatly affected our findings. And more importantly, a certain rate of misdiagnosis is a reality in daily clinical practice, so our findings are pragmatic in the sense that they apply to a real-life population of patients with both PD and parkinsonism. Another strength is that our study was based on highly standardized claims data. These data are a fair representation of the care provided, even though there is not a 100% match: mis-registrations or non-registrations can occur, as well as errors during the process of data registration by a healthcare professional or during the transfer of hospital data to the current database. A further strength is that we had a rather long follow-up relative to previous studies with a comparable approach. Finally, by using regression models, we did not have to divide our study sample into groups of lower or higher density values. This way, we avoided loss of data, making the analyses more accurate.

Our study also had some limitations, some of which relate to the generic limitations of observational studies of large data sets.(Bloem et al. 2018) For example, for primary care providers we were unable to determine if all claims were PD-specific, and their inclusion could lead to an overestimation of utilization and costs. Second, it was technically impossible to assess the duration of the disease when a patient had received the diagnosis before January 1st, 2008, which might have influenced our estimates for disease duration. A third limitation is that we faced many (arbitrary) decisions when

defining our methods. There are other models for assessing care density, for example, the model of Landon.(Landon et al. 2012) We preferred the method of Pollack et al. since it uses the patient's perspective, rather than the provider's perspective.(Pollack et al. 2012) Another methodological decision included the assignment of patients to a hospital, but not to other providers. For all such decisions, we tried to choose methods that had been used in previous research. However, given the limited PD-specific literature available for this topic, this was not always possible. A methodologic limitation is that not all providers in PD are always involved. When a less frequently involved provider is included in the patient's network, its density score will likely drop. However, this effect is partially mitigated by the inclusion of the independent variable "number of providers per patient", since less frequently involved providers will usually be involved in advanced stage of the disease when the more frequently involved providers are already present. Another limitation is that we did not analyze the characteristics of patients who were excluded from the analyses. We regarded them as data errors, or they might have received the diagnosis shortly before the end of the study period or they might have died just after the start of the study period. A final limitation is that our analyses did not adjust for correlations within hospital populations. However, we expect these correlations to be only small. This was corroborated by repeating the analyses using generalized estimating equations, which resulted in small estimated correlations between the residuals of patients within a hospital and little effect on the estimated coefficients in the models.

Our findings suggest that investing in the density of patient-sharing networks has the potential to increase the value of care for individual patients with PD. For individual healthcare providers, investing in density can be achieved by increasing the caseload of unique patients with PD, which in turn might lead to greater expertise. At the level of the patient population, this increased expertise might lead to better value of care and less medical practice variation. Such variations in care delivery are frequently reported in PD,(Bloem and Stocchi 2012; Willis et al. 2011; Dorsey, Vlaanderen, et al. 2016) but these are obviously unwanted as it leads to inequality in access to good care for different patients. And more importantly, a higher density was associated with better value of care and with better outcomes and lower costs. We therefore recommend that density scores be considered as a quality measure for network organizations. At the level of societies, density scores might act as a new tool for research on medical practice variation, or as an aid for contracting strategies of health care insurance companies. Further research might assess if this only applies to PD, or whether such an approach can also be extrapolated to other chronic neurological conditions.

APPENDIX 5.1 | **Odds ratios for logistic regression on health outcomes (adjusted)**

	Pneumonia			Orthopedic injuries			PD-related hospitalization			All PD-related complications			Mortality		
Parameter	**Estim.**	**95% C.I.**	**P-value**	**Estim.**	**95% C.I.**	**P-value**	**Estim.**	**95% C.I.**	**P-value**	**Estim.**	**95% C.I.**	**P-value**	**Estim.**	**95% C.I.**	**P-value**
(intercept)	0·113	0·083 - 0·154	<0·001	0·418	0·309 - 0·566	<0·001	0·666	0·456 - 0·973	0·036	0·778	0·556 - 1·090	0·144	0·000	0·000 - 0·000	<0·001
log(Density score)	0·926	0·889 - 0·964	<0·001	0·899	0·864 - 0·936	<0·001	1·023	0·971 - 1·079	0·392	0·901	0·862 - 0·941	<0·001	0·962	0·926 - 1·000	0·052
Age	1·028	1·024 - 1·032	<0·001	1·014	1·010 - 1·018	<0·001	0·964	0·960 - 0·969	<0·001	1·015	1·011 - 1·019	<0·001	1·129	1·124 - 1·135	<0·001
Sex (woman)	1·134	1·059 - 1·215	<0·001	1·520	1·417 - 1·630	<0·001	0·969	0·887 - 1·059	0·491	1·404	1·296 - 1·522	<0·001	0·665	0·621 - 0·713	<0·001
Duration of PD	0·971	0·921 - 1·023	0·271	1·063	1·009 - 1·120	0·022	1·421	1·318 - 1·533	<0·001	1·072	1·011 - 1·136	0·019	1·136	1·076 - 1·198	<0·001
PD in 2008	1·168	1·028 - 1·327	0·017	0·988	0·870 - 1·123	0·858	1·054	0·892 - 1·248	0·537	1·053	0·911 - 1·215	0·483	1·193	1·051 - 1·354	0·007
Number of providers per patient	1·014	1·000 - 1·027	0·042	1·064	1·049 - 1·080	<0·001	1·080	1·063 - 1·098	<0·001	1·074	1·056 - 1·093	<0·001	0·828	0·815 - 0·841	<0·001
Number of patients per provider	0·844	0·809 - 0·881	<0·001	0·931	0·893 - 0·970	0·001	0·983	0·928 - 1·040	0·547	0·888	0·849 - 0·929	<0·001	0·868	0·837 - 0·900	<0·001
AUC	0·601			0·606			0·671			0·616			0·788		
N	14,361			14,361			14,361			14,361			20,603		

APPENDIX 5.2 | **Parameter estimates for regression on log-transformed healthcare utilization**

	Number of neurologist visits			Number of physio-therapist visits			Number of occupational therapist visits			Number of speech & language therapist visits			Number of dietician visits			Number of psychologist visits		
Parameter	**Estim.**	**95% C.I.**	**P-value**	**Estim.**	**C.I.**	**P-value**	**Estim.**	**C.I.**	**P-value**	**Estim.**	**C.I.**	**P-value**	**Estim.**	**C.I.**	**P-value**	**Estim.**	**C.I.**	**P-value**
(intercept)	1·009	0·958 - 1·061	<0·001	1·150	1·046 - 1·254	<0·001	-0·313	-0·462 - -0·164	<0·001	0·678	0·450 - 0·906	<0·001	-1·038	-1·232 - -0·844	<0·001	-0·171	-0·362 - 0·019	0·078
log(Density score)	0·068	0·062 - 0·075	<0·001	0·052	0·038 - 0·065	<0·001	0·048	0·028 - 0·068	<0·001	0·024	-0·009 - 0·057	0·156	-0·013	-0·043 - 0·017	0·409	-0·032	-0·061 - -0·003	0·029
Age	-0·015	-0·015 - -0·014	<0·001	0·013	0·011 - 0·014	<0·001	0·013	0·011 - 0·014	<0·001	0·002	-0·001 - 0·005	0·143	0·015	0·013 - 0·018	<0·001	-0·004	-0·006 - -0·001	0·003
Sex (female)	-0·045	-0·057 - -0·033	<0·001	0·036	0·011 - 0·060	0·004	0·070	0·039 - 0·101	<0·001	-0·157	-0·209 - -0·105	<0·001	0·061	0·017 - 0·106	0·006	0·029	-0·020 - 0·079	0·243
Duration of PD	-0·140	-0·143 - -0·136	<0·001	-0·065	-0·072 - -0·057	<0·001	-0·115	-0·125 - -0·105	<0·001	-0·121	-0·137 - -0·105	<0·001	-0·145	-0·160 - -0·131	<0·001	-0·115	-0·131 - -0·100	<0·001
PD in 2008	0·370	0·351 - 0·390	<0·001	0·367	0·328 - 0·407	<0·001	0·382	0·333 - 0·431	<0·001	0·399	0·322 - 0·476	<0·001	0·426	0·357 - 0·496	<0·001	0·158	0·079 - 0·237	<0·001
Number of providers per patient	0·021	0·018 - 0·024	<0·001	0·179	0·173 - 0·184	<0·001	0·053	0·047 - 0·059	<0·001	0·049	0·039 - 0·058	<0·001	0·020	0·012 - 0·028	<0·001	-0·005	-0·015 - 0·005	0·347
Number of patients per provider	-0·010	-0·017 - -0·003	0·003	-0·175	-0·189 - -0·161	<0·001	-0·056	-0·078 - -0·035	<0·001	-0·041	-0·078 - -0·003	0·032	-0·041	-0·072 - -0·009	0·012	-0·014	-0·042 - 0·013	0·310
R^2	0·248			0·212			0·065			0·040			0·080			0·054		
N	33,703			33,474			14,534			8,895			6,490			6,437		

APPENDIX 5.3 | **Parameters of regression on log-transformed healthcare costs (adjusted)**

	Without costs for PD-related complication costs			With costs for PD-related complication costs		
Parameter	**Estim.**	**C.I.**	**P-value**	**Estim.**	**C.I.**	**P-value**
(intercept)	3·319	3·221 - 3·417	<0·001	3·539	3·443 - 3·635	<0·001
Log(Density score)	-0·018	-0·031 - -0·006	0·005	-0·030	-0·043 - -0·018	<0·001
Age	0·043	0·042 - 0·044	<0·001	0·043	0·041 - 0·044	<0·001
Sex (woman)	0·132	0·109 - 0·155	<0·001	0·133	0·110 - 0·155	<0·001
Duration of PD	0·032	0·025 - 0·039	<0·001	0·036	0·030 - 0·043	<0·001
PD in 2008	0·458	0·421 - 0·496	<0·001	0·413	0·377 - 0·450	<0·001
Number of providers per patient	0·015	0·010 - 0·020	<0·001	0·015	0·010 - 0·020	<0·001
Number of patients per provider	-0·075	-0·087 - -0·062	<0·001	-0·079	-0·092 - -0·067	<0·001
R^2	0·206			0·210		
N	35,414			35,414		

APPENDIX 5.4 | **Regression on the log-transformed density scores (adjusted)**

Parameter	**estimates**	**95% CI**	**P-value**
(intercept)	0·648	0·620-0·675	<0·001
% ParkinsonNet provider visits	1·164	1·145 – 1·184	<0·001
Number of providers per patient	0·005	0·002-0·009	0·004
Number of patients per provider	0·191	0·183 – 0·199	<0·001
R^2	0·325		
N	36639		

CHAPTER 6

Moving Parkinson care to the home

Dorsey ER, Vlaanderen FP, Engelen LJ, Kieburtz K, Zhu W, Biglan KM, Meinders MJ, Bloem BR. 'Moving Parkinson care to the home'. *Movement Disorders*, 2016; 31(9):1258-62.

ABSTRACT

In many ways, the care of people with Parkinson disease is poorly designed. Despite the documented benefits of receiving care from clinicians with Parkinson disease expertise, many (if not most) do not. Moreover, current care models frequently require older individuals with impaired mobility, cognition, and driving ability to be driven by overburden caregivers to large, complex urban medical centers.

Moving care to the patient's home would make Parkinson disease care more patient-centered. Demographic factors, including aging populations, and social factors, such as the splintering of the extended family, will increase the need for home-based care. Technological advances, especially the ability to assess and deliver care remotely, will enable the transition of care back to the home. However, despite its promise, this next generation of home-based care will have to overcome barriers, including outdated insurance models and a technological divide. Once addressed, home-based care will increase access to high quality care for the growing number of people with Parkinson disease.

6.1 THE SHORTCOMINGS OF CURRENT CARE

Current care models do not meet the needs of people with Parkinson disease (PD). (Bloem and Stocchi 2012; Willis et al. 2011; Achey et al. 2014) Many have limited access to proper care and that care, when delivered, is institution-based rather than patient-centered (Table 6.1). In this viewpoint, we examine the shortcomings in current care, the need for home-based care, the emerging models, and the barriers to overcome. While written from the perspective of the United States and Europe, the piece will hopefully have broader applications.

TABLE 6.1 | How different care models meet the needs of people with Parkinson disease

Feature	Individuals with Parkinson disease	Current care models	Home-based care
Location	Primarily suburban and rural areas (Kent 2015)	Primarily urban centers	Where the individual is located
Driving	Impaired ability (Santos-Garcia and de la Fuente-Fernandez 2015; Crizzle, Classen, and Uc 2012)	Usually requires driving	Little or no driving required (Qiang and Marras 2015)
Mobility	Limited(Parkinson 2002)	Generally required to access care	Not required to access care
Cognition	Frequently impaired (Aarsland et al. 2009)	Often demanding to navigate	Less demanding to receive
Disease course	Progressive (Parkinson 2002)	Least accessible for those with the most advanced disease	Accessible to those with greatest need (Dorsey et al. 2010)
Caregivers	Burdened (Martinez-Martin et al. 2007)	Increases the burden	Can reduce the burden

In 2001, the Institute of Medicine (now the Health and Medicine Division of the National Academy of Sciences, Engineering, and Medicine), a U.S. non-profit that provides independent, objective analysis, issued its landmark report *Crossing the Quality Chasm*. The report opens, "The American health care delivery system is in need of fundamental change." It further states, "Quality problems are everywhere, affecting many patients. Between the health care we have and the care we could have lives not just a gap, but a chasm."(Institute of Medicine (U.S.). Committee on Quality of Health Care in America. 2001) The report lists six aims to cross this chasm, proposing that health care should be safe, effective, patient-centered, timely, efficient, and equitable.

However, current care for PD, in the U.S., Europe, and likely the majority of the world, frequently does not meet these six aims. First, Parkinson disease care is often not safe. People with PD who are hospitalized are often subjected to delayed treatment, contra-indicated medications, prolonged immobility, lengthy stays, and high

mortality.(Gerlach, Winogrodzka, and Weber 2011; Aminoff et al. 2011) Second, while some comprehensive and distributed PD care models (Achey et al. 2014; Miyasaki et al. 2012) are quite effective, few patients receive such care, and many PD-related hospitalizations are likely preventable.(Willis et al. 2012) Third and fourth, providing patient-centered care that is timely has been studied little. (van der Eijk, Faber, Al Shamma, et al. 2011) Despite the limited evidence, focus groups and surveys suggest that people with PD want more personalized information from multiple disciplines that is delivered remotely in a timely manner.(van der Eijk, Faber, Al Shamma, et al. 2011; van der Eijk, Faber, et al. 2015) Fifth, PD care is very inefficient. Patients and their caregivers spend hours travelling and waiting in the clinic for routine follow-up appointments.(Dorsey et al. 2013) Outside the clinic, nearly 25% of Americans over 65 with PD reside in expensive nursing homes that cost more than $200 per day(Safarpour et al. 2015) – money than could be devoted to preventing the need for institutional care for many.

Finally, perhaps most concerning is the inequity of current PD care. A primary determinant of care received remains where you live. In the U.S., 42% of people with PD over 65 and up to 100% of individuals in some rural areas do not see a neurologist soon after diagnosis.(Willis et al. 2011) In Europe, the first right expressed in the European Parkinson's Disease Association Charter is care from a physician with a special interest in PD,('The European Parkinson's disease Standards of Care Consensus Statement' 2011) yet 44% of Europeans do not see a PD specialist in the first two years after diagnosis.(Bloem and Stocchi 2012) Beyond neurological care, access to specialist nurses, occupational therapists, and counselors is often more limited.(Stocchi and Bloem 2013) In less wealthy countries, the situation is even worse. China only has approximately 50 movement disorder specialists to care for over two million people with PD.(Dorsey and Willis 2013) In Bolivia, a door to door epidemiology study found that none of the persons identified with PD had ever seen a physician, much less received treatment.(Nicoletti et al. 2003)

New, comprehensive PD care models are emerging that seek to deliver care that is aligned with the aims articulated by the Institute of Medicine. (Achey et al. 2014; van der Marck, Bloem, et al. 2013) For example, ParkinsonNet, a comprehensive, multi-disciplinary care model in the Netherlands, can enhance care, improve health, and lower costs.(Achey, Aldred, Aljehani, Bloem, Biglan, Chan, Cubo, Dorsey, Goetz, Guttman, Hassan, Khandhar, Mari, Spindler, Tanner, van den Haak, Walker, and Wilkinson 2014; van der Marck, Bloem, et al. 2013) Still, these models are resource intensive and demanding of patients and their caregivers. For example, integrated multi-disciplinary care models require individuals with limited mobility and driving ability to visit physical therapists and speech therapists three times per week for several weeks. Surgical treatments, such as deep brain stimulation, make similar

requirements – often lifelong ones – for follow-up care for individuals with more advanced disease.

6.2 NEED FOR HOME-BASED CARE

To improve PD care, more of it must be delivered at home. Home care is not new. In the early part of the 20th century, the house call was a dominant means of providing care, with 40% of physician-patient encounters in 1930 occurring in the home. (Meyer and Gibbons 1997) However, advances in transportation and diagnostics that had to be delivered in medical centers (e.g., x-ray, EKG) contributed to the house call's decline. Early in the 21st century, house calls are returning. They are available through home-based chronic care models,(Landers 2010) on-demand house calls by physicians for episodic care,(Jolly 2015) and a "hospital at home" model.(Cryer et al. 2012) The latter model provides hospital-level care, including physician and nurse visits and intravenous medications, for acute conditions like pneumonia directly in the home. These models can generate equal or better clinical outcomes, improve patient satisfaction, and lower costs, (Cryer et al. 2012) yet few have been applied to PD. In addition to in-person care at home, the next generation house call, enabled by advances in telecommunications, is also emerging.(Dorsey et al. 2013; Achey et al. 2014) Through video visits, these virtual house calls enable frequent consultations and provide specialty care to patients independent of geography.

The demand for in-home care is likely to grow due to demographic, social, and technological factors. Both the absolute number and proportion of older people with PD will increase. Due to aging populations, the prevalence of PD in the world's most populous nations will rise to over 8.7 million patients, twice as much as it was in 2005. (Dorsey et al. 2007) Similarly, rising life expectancies (four years in the U.S. and six years globally in the last two decades)("Health Care's Big Spenders: The Characteristics Behind the Curve" 2016) along with new therapies for advanced PD(Olanow et al. 2014) may increase the survival of people with the condition, leading to more people with advanced disease.

Social shifts are also driving home care. The splintering of the extended family, the increased mobility of the nuclear family, and the strong desire of older individuals to remain in their own homes(Levitz) lead to geographically separated children caring for aging parents. These children will increasingly demand technology solutions that enable them to care for their parents, monitor their health, and connect to their parents' clinicians conveniently. In addition, more older individuals are discovering the internet, tablets, and smartphones for themselves.(Perrin and Duggan June 26, 2015).

Lastly, technological advances are enabling people with PD and other chronic conditions(Darkins et al. 2008) to access specialists in satellite clinics or in their homes.(Achey et al. 2014) Preliminary evidence suggests that web-based video conferencing may offer similar clinical benefits to that of in-person care while saving patients and caregivers 100 miles of travel and 3 hours of time per visit.(Dorsey et al. 2013) Multidisciplinary care, including speech therapy,(Constantinescu et al. 2011) mental health care, and "tele-rehabilitation"(Bloem, de Vries, and Ebersbach 2015) can also be delivered remotely.

6.3 EMERGING HOME-BASED CARE MODELS

Emerging care models will combine remote monitoring, self-monitoring, and multi-disciplinary care to enable the provision of patient-centered care at home and decrease the need for in-clinic assessments. Remote monitoring from devices, such as wearable sensors,(Ferreira et al. 2015) smart beds, wall-mounted cameras, smart glasses, and even utensils, can monitor a patient's symptoms and function objectively in their environment, facilitating the delivery of highly personalized care. (Espay et al. 2016; Maetzler et al. 2013) These devices increasingly form the "Internet of Things,"(Pasluosta et al. 2015) a network of objects that can collect and exchange data, and can measure relevant outcomes (e.g., physical activity, sleep, falls) that are hard to assess using traditional questionnaires or personal interviews.

Issues such as feasibility (can patients manage these new devices?), compliance (can patients handle prolonged use of wearable sensors?), and validity (do the devices capture clinically relevant information that inform care?) remain to be addressed, but the initial experience is positive, provided that patients are fully informed and engaged from the outset.(Ferreira et al. 2015) Wearable devices will also shift current snapshot measurements in the clinic into a more constant flow of measurements in the comfort of the patient's own surroundings, allowing for more ecologically valid observations.

In addition, the increasing ubiquity of smartphones is enabling self-management by patients through self-monitoring apps.(Arora et al. 2015) These apps allow patients to record symptoms and signs, track progression, and identify warning signals that may necessitate a clinical follow-up. When integrated with the hospital-based electronic health record, these data will provide feedback to the clinician to guide treatment decisions and improve health outcomes. When combined with online education, such as a web-based, informative, and interactive television program (e.g., www.ParkinsonTV.nl) or social media-based community building,(Achey et al. 2014) remote monitoring tools can increase the ability of people with PD "to adapt and self-manage," a new definition of health.(Huber et al. 2011)

Finally, multidisciplinary care both in-person and remotely can be delivered into the home. The combination of in-person consultations in the home (e.g., to develop personal relationships or to conduct detailed examinations) and remote consultations in the home (e.g., to provide ongoing care) could meet the needs of patients.(Qiang and Marras 2015) Such a combination of in-person and virtual house calls can reveal information that is not easily observed in clinic, where patients often perform very differently compared to their usual behavior at home. These house calls can also provide valuable insights into a patient's domestic circumstances (e.g., safety of physical environment, level of social support). Specialized Parkinson's nurses, who have a broad perspective and can act as liaison to other healthcare professionals within the team, have already begun offering house calls.(Jolly 2015) In an ideal situation, such Parkinson's nurses could leave the clinic as agents of integrated regional networks of professionals specialized in PD. House calls are also important for other PD professionals, including physiotherapists (e.g., to help patients learn to transfer from their own beds) and occupational therapists (e.g., to remove domestic hazards). Current professional guidelines, for example, recommend that certain assessments (e.g., transfers) are best done at home. (Keus et al. 2014) Finally, some remote care could be delivered asynchronously with information flowing from patients to clinicians and advice being delivered from clinicians to patients or other clinicians.(Cubo ; Wilson and Maeder 2015)

6.4 OVERCOMING BARRIERS

Several barriers, including reimbursement, access to technology, and limited evidence, can slow the migration of care to the home. Currently, major insurers, including Medicare (the universal health insurer for older Americans), incent institution-based care by paying more for care rendered in institutions than in the home. Organizations that have integrated delivery and financing of health care can benefit from cost savings from home-based care and thus are likely to be early adopters of this patient-centered model. In the U.S., the Department of Veterans Affairs(Darkins et al. 2008) and Medicare Advantage programs(Tompkins C 2013) have implemented home-based care models. Kaiser Permanente, a large integrated health system in the U.S., uses internet, mobile, and, more recently, video technology to improve outcomes and increase convenience for its patients.(Pearl 2014; Vlaanderen et al. 2016) In the Netherlands, ParkinsonNet is developing an integrated reimbursement system to assist their network approach of care delivery, which includes many telehealth solutions as video consultations and online platforms for patients. Countries with single payer health systems, like Canada, Norway, or Luxemburg, are also poised to realize the advantages of home-based care for PD. Likewise, across Europe, consumer choice and flexibility have become a major goal of modern home-based care systems.(Colombo and Organisation for Economic Co-operation and Development. 2011) However, funding for these systems remains a

challenge because hospitals lose income in a “fee-for-service” model by facilitating care outside the hospital.

The digital divide,(Norris 2001) the differential access to internet and telecommunication technologies based on economic and social factors, prevents the use of technology to receive care at home. People who are older and have more chronic conditions are less likely to use the internet,(Smith 2014; Fox and Purcell 2010) and the digital divide has hampered efforts to use technology to deliver PD care at home. (Dorsey, Achey, et al. 2016) The divide can be overcome by delivering in-person home care to people with PD,(Hack et al. 2015) providing remote care via satellite clinics close to one’s home as is done in Canada,(Achey et al. 2014; Qiang and Marras 2015) engaging children in the care of their parents, and increasing access to telecommunication technologies. The digital divide is narrowing and the increasing ubiquity of smartphones, which are projected to be in the hands of 90% of individuals over age 6 by 2020,(Dorsey, Vlaanderen, et al. 2016) provides a promising avenue to increase access to care, especially in resource limited countries like China and India.(Tian et al. 2015)

Despite its promise, evidence for these new, home-based care programs is needed. Some of that evidence is being gathered currently,(Dorsey, Achey, et al. 2016) and some is being generated in other chronic conditions.(Power and Ashby 2014; Landers 2010) Preliminary interest in these models for PD is robust. For example, over 11,000 individuals from 80 countries and all 50 U.S. states visited a recruitment website for a randomized controlled trial of virtual house calls for PD.(Dorsey, Achey, et al. 2016)

Notwithstanding the barriers, “[it] seems inevitable that health care is going home.” (Landers 2010) Ushering in the next generation of home care will require collective efforts from patients, families, clinicians, advocates, philanthropists, insurers, technology firms, and policy makers. Unless these models gain more visi-bility, these stakeholders will remain ignorant of opportunities to develop, fund, evaluate, and advocate for models to improve care. To bridge the chasm identified by the Institute of Medicine, we need to be more critical of our current care models, more willing to experiment with disruptive ones, and more prepared to refine them. The more clearly we envision and adopt these future models, the sooner the growing number of people with PD will realize their benefits.

CHAPTER 7

General discussion

In this chapter, I discuss the main findings of this thesis and combine them to construct general conclusions. I start with an overview of the findings of the five individual chapters (paragraph 7.1), and then discuss the interpretations of these findings and how they relate to each other (paragraph 7.2). I then point out the strengths and limitations of our work (paragraph 7.3) before formulating our conclusions and presenting an outlook to the future (paragraph 7.4).

7.1 OVERVIEW OF THE MAIN FINDINGS

Parkinson's disease (PD) is a progressive, disabling chronic condition. Its management does not only require treatment by knowledgeable professionals, but also seamless and sustainable care provision. Seamless care is understood to be *"care which is consistent and coherent, marked by an orderly, logically and aesthetically consistent relation of parts, without discontinuities or disparities, uniform in quality and combined in an inconspicuous way"* (Hammond 2010). Sustainable care is defined as *"care that can be maintained through time, and that does not drain ending resources, most notably financial boundaries"* (Jeurissen, Maarse, and Tanke 2018). However, in my introduction chapter **(Chapter 1)**, I stated that care for people with PD is often neither seamless nor sustainable. In the work presented in this dissertation, I aimed to answer the critical and vexing question how to best achieve seamless and sustainable care for people with PD.

I first reconstructed, from a national administrative medical claims database, a sex-specific patient journey for Dutch people with PD during the first 5 years after diagnosis **(Chapter 2)**. I included claims data of 13,518 men and 8,775 women with newly diagnosed PD. The reconstruction revealed quantitative information about healthcare utilization and the occurrence of clinical milestones over time. It also revealed profound sex differences: after diagnosis, women visited on average their general practitioner 10 days and physiotherapist 89 days sooner than did men. I found little difference in neurologist utilization. Approximately two years after diagnosis, the first PD-related complication occurred (women: 1.8 years; men: 2.3 years). After five years, 37.9% of the women had visited an occupational therapist and 18.5% a speech & language therapist at least once. The corresponding figures for men were 33.1% and 23.7%. Moreover, 72.9% of women and 68.7% of men had experienced at least one PD-related complication, such as pneumonias, orthopaedic injuries and unexpected hospital admissions; 27.5% of women and 22.5% of men were admitted to a nursing home. Within five years after diagnosis, 14.6% of women and 18.3% of men had died. The identified sex differences contribute to the debate about phenotypical differences in PD between men and women. Insight into the sex differences might also lead to better patient-centred care.

To identify seams in healthcare from the perspective of people with PD, the Voice of the Customer approach was applied: a methodology originally developed in industry to probe for the clients' needs **(Chapter 3)**. The participants reported unmet needs mainly concerning the social, emotional or domestic domain, rather than the bio-medical aspects of the disease. Their top unmet needs were: (1) more self management; (2) better interdisciplinary collaboration between different healthcare providers; (3) more time to discuss the future and possible scenarios; and (4) one healthcare provider acting as case manager, either to solve problems directly or to direct a person with PD to the healthcare provider best equipped to address the problem at hand.

Outcome-based payment models (OBPMs) might contribute to seamless and sustainable care for people with PD by aligning incentives with quality of care, rather than with volume of care. Regrettably, no specific OBPM has been designed, implemented and evaluated for PD care. I therefore reviewed the effects of OBPMs in general healthcare on quality of care and healthcare costs **(Chapter 4)**. Our review provided an analysis of the characteristics and effectiveness of OBPMs, enabling to identify models that lead to favourable effects. 88 studies could be included, describing 12 OBPMs. Based on differences in design features, two groups were distinguished: narrow OBPMs, which only contain financial incentives for objectively measured quality performance; and broad OBPMs, which combine global budgets and risk sharing for multidisciplinary provider groups with financial incentives for quality. Five out of nine narrow OBPMs showed positive effects on quality; the others had mixed (2) or negative (2) effects. The effects of narrow OBPMs on healthcare utilization or costs were, however, either unfavourable (3) or unknown (6). All broad OBPMs (3) showed positive effects on quality of care, while reducing healthcare cost growth.

Although only three thoroughly evaluated, broad OBPMs could be included in this review, their effects on both quality of care and healthcare utilization/costs are more favourable as compared to the narrow OBPMs. Moreover, the effects of the broad OBPMs improved over time, whereas the effects of narrow OBPMs tended to be more short-lived. I also found that in both groups of OBPMs the process indicators showed larger improvements than did the outcome indicators. Other findings were: (1) larger private providers and providers with initially poor quality scores tended to score better than other providers; (2) high-need patients do not seem to benefit more from OBPMs than other patients; (3) broad OBPMs had little effect on non-incentivized indicators, while there are signs that non-incentivized indicators may deteriorate in the narrow OBPMs; (4) narrow OBPMs do not seem to decrease social or ethnic disparities; and (5) narrow OBPMs do not seem to lead to gaming on a large scale.

Every person with PD receives care from multiple healthcare providers, who form what is known as a 'patient-sharing network'. These networks differ in density: the

number of mutual people with PD treated by the network members. I assessed whether denser patient-sharing networks can be linked to better health outcomes, lower healthcare utilization and lower healthcare costs **(Chapter 5)**. To quantify density, the density model of Pollack et al was used (Pollack et al. 2012), which calculates a density score: the more mutual patients are served by a patient-sharing network, the higher the network's density score. density scores differed on the patient level, but also on the level of hospital populations. The average density score per patient was 6.7 with a standard deviation of 8.2. The top three hospital populations had average density scores of, respectively, 8.8, 8.3, and 8.0; the bottom three had, respectively, 2.1, 2.0, and 1.6. Adjusted for confounders, denser networks were found associated with lower occurrence of PD-related complications (OR: 0.901; $p<0.001$), especially pneumonias and orthopaedic injuries, but not associated with lower mortality (OR: 0.962; $p=0.052$). Higher density scores were associated with higher utilization of neurologists, physiotherapists and occupational therapists (coefficients: 0.068, 0.052 and 0.048, respectively; p-values all <0.001) and lower utilization of psychologist (coefficient: -0.032, $p=0.029$) and lower healthcare costs (coefficients: -0.018, $p=0.005$ and -0.030, $p<0.001$ for with and without costs for complications, respectively). A secondary analysis showed that a strong correlation between the density score and the percentage of visits to providers who are members of ParkinsonNet (a Dutch not-for-profit organization aiming to improve quality of care for people with PDs; see also Table 1.2). This suggests that the effects might in part be caused or influenced by the efforts of ParkinsonNet.

In **Chapter 6**, I discussed how we could improve the care for people with PD by moving it to the patient's home. Care of people with PD is poorly designed. For one thing, despite the documented benefits of receiving care from clinicians with PD expertise, many people with PD do not receive this expert care. Moreover, current care models frequently require older individuals with impaired mobility, cognition, and driving ability to be driven by overburdened informal caregivers to large, complex urban medical centres. Moving care to the patient's home would make PD care more patient-centred. Demographic factors, including aging of the populations and other social factors, such as the splintering of the extended family unit, will increase the need for home-based care. Technological advances, especially the ability to remotely assess and deliver care, will enable the transition of care back to the home. However, despite its promise, this next generation of home-based care will have to overcome barriers, including outdated insurance models and a technological divide. Once addressed, home-based care will increase access to high-quality care for the growing number of people with PD.

7.2 INTERPRETATION

In this paragraph, I provide interpretative answers and discuss possible approaches on how we can best achieve seamless and sustainable care for people with PD.

What are the current seams in the care for people with PD and how can we identify them?
Care for people with PDs in the Netherlands (and in many other countries in the world) is currently not seamless. Earlier work reported delays in the diagnostic process or the diagnosis delivery (Bloem and Stocchi 2012), difficulties in finding allied healthcare professionals or specialized nurses (Stocchi and Bloem 2013), limited information provision, and a lack of multidisciplinary collaboration (Bloem and Stocchi 2015; van der Eijk, Faber, Al Shammaa, et al. 2011; Buetow et al. 2008). My present findings align with these studies and add some further important 'seams', such as the great variation in the care that different people with PD receive. This suggests that not all people with PD receive the support from healthcare providers that they need and deserve. A remarkable finding is that women receive care earlier after their diagnosis than men **(Chapter 2)**, even though PD progresses at an equal or even faster rate in men. I also found considerable variation in the density of provider networks. This variation in density can affect the quality of care and health outcomes **(Chapter 5)**. Additionally, there is a substantial amount of variation in healthcare use across people with PD as well **(Chapters 2 and 5)**. PD is more common among men than women, by a ratio of roughly 60%:40% (as was confirmed in Chapters 2 and 5). However, the total healthcare costs per year for PD are equally divided over the sexes (National Institute of Public Health and the Environment 2019c). For residential care, the total costs for women are significantly higher than for men (National Institute of Public Health and the Environment 2019c). This implies that women face higher per capita costs than men. Despite some reasons why care delivery might slightly differ between men and women (Göttgens et al. 2020), they are likely to be treated according to the same guidelines. Therefore, the afore mentioned variations might be signs that women either receive unnecessary care, or that their care is delivered too late or in an inadequate fashion, leading to disease complications that are both costly and perhaps partially avoidable.

The seams identified above all concern organisational aspects, rather than bio-medical aspects: in **Chapter 3**, respondents had not encountered problems with access to care, either financially or otherwise, or seams regarding medication and therapies. The most urgent seams reported by the people with PD concerned one's social, emotional or domestic domain. The respondents wished for more self management; more emotional support, and more time to discuss future scenarios. Therefore, although healthcare providers do very well in the medical aspect of treating PD, they perhaps do not pay enough attention to how people with PD experience this care. Importantly, they also dearly missed a single point of access, i.e. one healthcare provider acting as

a personal case manager who is readily accessible for answering simple questions (**Chapter 3**), or a few providers who share the responsibility for navigating people with PD to the appropriate healthcare provider (van Halteren et al. 2020).

To identify seams in care, we need to include the perspective of the individual patient **(Chapter 3)**. This is often achieved through open or semi-structured interviews, focus group interviews or questionnaires. These are methods that tend to focus on the biomedical domain or the professional-patient relation. I introduced a new interview method to assess unmet patient needs, named the Voice of the Customer method **(Chapter 3)**. This method places the interviewee in the expert role while being in his or her own environment. This creates an atmosphere in which the interviewee feels safe to share his or her deepest feelings. The social, emotional and domestic domains might be more accessible by using such method **(Chapter 3)**. When aiming for a complete inclusion of seams in care from the patient's perspective, the Voice of the Customer method might be a welcome contribution to further research, as we continue our quest to improve the quality of care for people living with PD.

How can we make care delivery for PD more seamless and sustainable?
Combining the results of the individual chapters brings us to three main suggestions to address the identified seams. First, the way in which healthcare providers are organised and have divided tasks needs to be rethought. When the disease progresses, complications related to PD are likely to occur, and progressively more different healthcare providers may get involved **(Chapter 2)**, necessitating multidisciplinary network approaches. The patient, however, may have difficulty to oversee the situation when the number of involved healthcare providers is increasing. When this moment arrives (but perhaps even earlier in the disease process), it would be wise to appoint one of the healthcare providers as the personal case manager **(Chapter 3)**, or organise case management among few providers with shared responsibilities in different domains of care (van Halteren et al. 2020).

Theoretically, any member of the multidisciplinary team could take on this task of personal case manager, as long as it is clear who is now responsible for this specific task, and of course, provided that sufficient time is available to fulfil these responsibilities. Having said that, I do think that specialist Parkinson's nurses might be the most suited for this task. In the Netherlands, they already play a central role in care delivery, especially the care delivered in the hospital (Bloem et al. 2010). If a hospital employs a specialist Parkinson's nurse, almost all patients are seen by this nurse several times a year (Bloem et al. 2010). Moreover, compared to the consultant neurologist, they are usually more readily accessible for people with PD, for example by phone or email. Challenges might arise in the social and domestic domains, where these nurses are normally less involved, which are perhaps not necessarily the responsibility of the

hospital-based medical team, but which are presumably more appropriate areas to be addressed by general practitioners or other community-based professionals. Alternatively, home visits by Parkinson nurse specialists to discuss any problems in these domains could be helpful. Ideally, these specialist nurses would leave the clinic as an agent of the integrated regional network of healthcare providers specialized in PD **(Chapter 6)**. They should not only arrange care for their patients, but also help them finding their way in the world of long-term care and welfare, and provide emotional support. Such case managers have been successfully and sustainably implemented in the care for people with other neurodegenerative disorders (MacNeil Vroomen et al. 2016; Alzheimer Nederland 2020), but there is still relatively little experience with this in the field of PD. Interestingly, two recent studies have examined the value of home visits, in one study performed by just the Parkinson nurse (Eggers et al. 2018), in another study by the nurse combined with other members of the multidisciplinary team (Fleisher et al. 2018). Currently efforts are made to examine if such interventions are cost-effective (Radder et al. 2020).

Second, technological innovations such as video visits and self-monitoring apps can help to mend some seams in PD care, at least in part (Mancini et al. 2020). Remote monitoring from smartphones and devices such as wearable sensors, smart beds, wall-mounted cameras, smart glasses, and digitised utensils may help to move clinical measurements back into the patient's home. These innovations hold promise of making the retrieved values more valid **(Chapter 6)**. However, the digital divide (Norris 2001), with differential access to internet and telecommunication technologies, may prevent the use of technology to receive care at home for a sizeable proportion of people with PD in the world (Smith 2014; Fox and Purcell 2010; Dorsey, Achey, et al. 2016). This divide can be overcome by delivering in-person home care (Hack et al. 2015), providing remote care via satellite clinics close to one's home (Achey et al. 2014; Qiang and Marras 2015), engaging children in the care of their parents, and improving access to telecommunication technologies (Chapter 6).

Finally, there is a need for new, home-based care programs. Currently, much of the care for people with PD is delivered at the hospital, and many treatment decisions are made based on the relatively brief consultations there. This creates difficulties for people with PD living far away from their hospital **(Chapter 6)**. Additionally, it may be hard for healthcare providers to capture a patient's home situation during these brief consultations (Bloem et al. 2020). To overcome this, care needs to be delivered more closely to, or ideally within, the patient's own home **(Chapter 6)**. One way is to intermittently use both virtual house calls and in-person consultations, where the relative proportion of these two types of consultations should be dictated by the individual preferences and clinical needs of each person living with PD (Mancini et al. 2020). This individually tailored approach would be an attractive way to optimally

meet the needs of people with PD, while also improving their clinical outcomes, daily functioning and quality of life, and without increasing social and healthcare costs (Qiang and Marras 2015). It might also offer people with PD a greater span of control over their own health. Being more skilled in self-management was one of the main identified needs from **Chapter 3**, in line with previous work as well (Bloem and Stocchi 2015). Digital or in-person home visits also help healthcare providers to obtain a better impression of how their patients are functioning at home, rather than in the clinic where many people with PD perform paradoxically rather well. Freezing of gait, for example, is notoriously difficult to elicit during in-person visits to the hospital, even among people who are severely debilitated by very regular episodes of marked freezing at home. People with PD then have the opportunity to show their healthcare providers what problems they experience in their daily lives, and providers can more easily suggest individually tailored solutions.

How should we pay for care for people with PD in order to be more seamless and sustainable?

Traditional fee-for-service payment models have important shortcomings. They incentivise volume, thus increasing healthcare costs, while lacking an incentive for improving quality of care (Orszag and Ellis 2007; Tai, Kalanithi, and Milstein 2014; Jeurissen, Maarse, and Tanke 2018). As shown in **Chapter 4**, outcome-based payment models (OBPMs) might be a good alternative to counter these shortcomings. OBPMs are payment models that align financial incentives with outcomes and quality of care **(Chapter 4)**. An OBPM for PD was not yet available, but PD shares many features with other chronic diseases that have been addressed successfully using these models **(Chapter 4)**. As is the case for many other chronic conditions, optimal management of PD requires a multidisciplinary team approach **(Chapters 2, 3, 5 and 6)**. However, this multidisciplinary care approach is often poorly organised **(Chapter 6)** with great variation in care delivery (Chapter 2), and does not meet the expectations of people with PD **(Chapter 3)**.

Of the two identified types of OBPMs in **Chapter 4**, 'broad' OBPMs, which combine global budgets and risk sharing with financial incentives for quality, had the most promising results. Unlike the 'narrow' OBPMs, which contain financial incentives for quality measures alone, broad OBPMs have shown the ability to save costs over time (Eijkenaar and Schut 2015; Song et al. 2011; Song et al. 2012; Song et al. 2014; McWilliams, Landon, and Chernew 2013; McCarthy 2015) **(Chapter 4)**. Furthermore, broad OBPMs seem to be less prone to ceiling effects (Eijkenaar and Schut 2015; Song et al. 2011; Song et al. 2012; Song et al. 2014) **(Chapter 4)**. Another disadvantage of narrow OBPMs is that non-incentivised indicators may deteriorate (Calikoglu, Murray, and Feeney 2012; Doran, Fullwood, et al. 2008; Campbell et al. 2009) (Chapter 4). This is much less seen in broad OBPMs (Chien et al. 2014; McWilliams, Landon, and Chernew

2013) possibly due to the additional incentives for cost containment in broad OBPMs (i.e. global budgets and risk sharing). These incentives create an extra incentive to minimize avoidable healthcare use due to sub optimal care **(Chapter 4)**. Broad OBPMs also seem to be better applicable in PD due to the nature of the provided care. In general, people with PD receive care from a hospital combined with care from multiple primary care providers. **(Chapters 2 and 5)**. The global budgets of OBPMs can target the complete chain of delivered care, whereas narrow OBPMs typically target specific institutions.

I would therefore welcome the development and subsequent rigorous evaluation of an experimental broad OBPM that links financial incentives to quality indicators relevant for PD. As a pilot we would suggest a model as shown in Figure 7.1. This models is based on features of broad OMPMs (global budgets, different providers, shared risks; see **Chapter 4**), while being disease-specific (a feature of narrow OBPMs; **Chapter 4**). Depending on the existing healthcare system, target populations could be all people with PD of a specific geographical area, or all insured people with PD of a particular insurance company. The first option seems more applicable in a single payer system; the second in a multiple payer system. Benefits from shared risks should be dependent on quality requirements. These requirements should include outcome indicators of PD-specific complications that are common, debilitating and costly, such as aspiration pneumonias and fractures of the hip or elsewhere due to falls, or avoidable PD-related hospitalisations. Risk mitigating measures for providers are essential to ensure reliable data and prevent providers from 'gaming' the model: selective inclusion or manipulation of the data in order to maximise financial gains. The incentives should be combined with a global budget for PD care, with shared risks for providers and healthcare payers. A shown in **Chapter 4**, this also improves the sustainability of these models. The budgets might be attributed to healthcare organisations or geographical regions.

Further selective attribution of these budgets would concentrate the care for people with PD among fewer providers. This might not only be a stimulus for more efficient care delivery, but also should be used to increase the density of care; this is a measure of the number of mutual patients that the different providers share (Pollack et al. 2012). Dense providers might communicate and cooperate better (Foy et al. 2010; Barnett et al. 2011), or know each other better via referrals (Barnett et al. 2011). This might lead to better coordination and organization of care for their patients (Pollack et al. 2012). As we have seen, care for people with PD becomes more seamless and more sustainable if there is a high density between the different providers of care **(Chapter 5)**. Density itself could therefore act as a quality indicator for the payment model, comparable with current volume standards in surgical procedures.

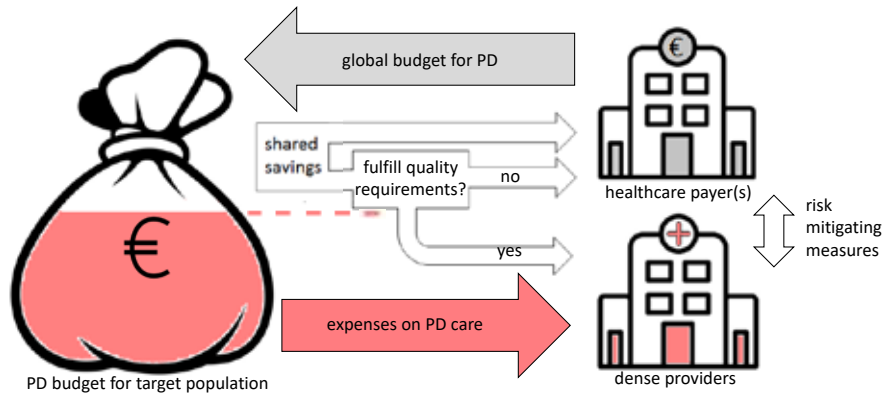

FIGURE 7.1 | **the suggested outcome-based payment model for PD**

However, our methods of studying non-PD-specific OBPMs contain limitations which make it still uncertain if such an OBPM will be a success. First, comparing different outcome measures, used in different OBPMs, is not ideal. Some outcome indicators may have more improvement potential than others, and the availability of clear guidelines can increase this potential. Second, our review included OBPMs from both the inpatient and the outpatient sectors, which operate differently and can be subject to different payment and billing systems. Third, the effects of the payment models are likely to be influenced by contextual factors, and most models in this review stem from the USA. Extrapolation of these findings from USA-based studies to other healthcare systems (and to PD: a different disease) is hard. Fourth and finally, our suggested model is disease-specific which aids to the conciseness of the pilot. However, this counts as a feature of narrow OBPMs and might affect few patients per provider. We hope this pilot might be extended to other chronic diseases or can be incorporated in other broad OBPM initiatives. Together with robust risk-mitigating measures, this should diminish the incentives for gaming that accompany small numbers of patients per provider.

The aim for higher density might have disadvantages as well. A higher density might lead to concentration of care. This limits the number of providers that a person with PD can choose from, thus obstructing their ability to freely choose a desired provider. However, having a dense specialised network could still coincide with the ability of patients to choose a different provider who operates largely in isolation outside the network, provided that the patient receives clear upfront information about the expertise that each provider can offer, and about the outcomes that are likely to occur when being treated by either an expert in a network or an isolated provider outside

the network. Freedom of choice is important, but should be based on transparent information about the offerings of each provider. Having more dense networks might also increase the distances that people with PD may need to travel in case they require an in-person assessment. At the same time, as I discussed earlier, the need for such in-person visits for chronic follow-ups may diminish as the opportunities for telemedicine services continue to improve. Also, the willingness of people with PD to travel longer distances will likely depend on the received urgency of the problem at hand; many patients are typically to travel longer distances to receive the best possible care for important and vexing clinical issues. And finally, higher density and concentration of care can cause increased market power for the involved healthcare providers, leading to higher prices. Further research should assess if this is the case for PD, and if density has the same effects on the treatment of other chronic conditions.

7.3 STRENGTHS AND LIMITATIONS

My methods had several strengths and limitations. The methodological limitations of each research project have been addressed in the designated chapters. However, several additional strengths and limitations need to be mentioned regarding the interpretation of our findings in relation to the research questions.

Strengths

One important strength is that our research applied a mix of research methodologies. **Chapters 2 and 4** consist of quantitative database analyses of data of large samples of people with PD (22,293 in **Chapter 2** and 50,508 in **Chapter 4**). These samples constitute de facto the whole patient population of the Netherlands. The analyses were driven by a predefined set of analyses, rather than an exploratory approach. The two empirical studies are supplemented with a qualitative study and a provocative viewpoint. Additionally, the findings of **Chapter 4** are from a comprehensive systematic review consisting of 12 different OBPMs, retrieved from four different data sources.

Limitations

Several limitations need to be mentioned. An important limitation concerns the assessment of the current state of the PD care by reconstructing the gender-specific patient journey **(Chapter 2)**. Although the conclusions of this study are based on a large sample, the sample consisted only of people with PD who were in their first five years of their disease. The state of care for people with PD with a longer disease duration could therefore not be assessed. This is an important limitation, since PD is a progressive disease, and people with PD over time require more, and more complex, care from an increasing number of professional disciplines (Oliver et al. 2019; Fleisher et al. 2019; Spindler et al. 2019). A similar approach for people with PD with longer disease duration would require a database that can go further back in time than ours.

A second limitation is that our review of OBPMs **(Chapter 4)** is not PD-specific. All included models covered a range of chronic conditions or entire healthcare sectors, such as primary care. None of these OBPMs is disease-specific, let alone PD-specific. Drawing conclusions about the effects of these payment models on quality of care and healthcare costs for PD might therefore be a little far-fetched. However, the results of these models showed especially improvements in managing chronic conditions. I do not have reasons to think PD might be an exception to this, due to its chronic character and the availability of clear outcome measures.

Finally, the activities of the Dutch ParkinsonNet might have influenced the generalisability of our findings in **Chapter 3 and 5.** ParkinsonNet is a Dutch network of over 3,600 specifically trained healthcare providers who strive to improve multi-disciplinary collaboration, increase PD-specific knowledge, and stimulate patient-centeredness. The interviewed people with PD in **Chapter 3** were all from the area of Nijmegen, the Netherlands, a region in which ParkinsonNet deploys relatively many activities and experiments. Therefore, it might be possible that the identified needs are not equal to those in regions of the Netherlands where ParkinsonNet is less active, or in other countries where such a professional network approach is not operational at all. As already stated in **Chapter 5**, ParkinsonNet actively influences the density of provider networks for people with PD, while its other activities (providing education, developing guidelines) influence quality of care too. Additionally, as stated in **Chapter 6**, many countries lack organizations attempting to improve the care for people with PD.

7.4 CONCLUSIONS AND FUTURE OUTLOOK

I draw the following conclusions and suggestions (Box 7.1). First, PD requires a multidisciplinary treatment approach by professionals working in different echelons of healthcare, which provides great challenges to the current organisation of care. We should aim for care that is more seamless and sustainable.

Second, women with PD receive more and earlier healthcare across almost all disciplines. I expect that this might be due to a doctor delay, a patient delay, or both in the diagnostic process in women. Maybe care could become more seamless if healthcare providers would be more alert with respect to the early signs of PD in women. I would therefore encourage more research on differences in health care utilisation between men and women living with PD. There is ample literature about differences in disease presentation or progression between the sexes. What is missing is research on possible sex-specific delays on behalf of doctors or patients in the management of PD. I believe that more knowledge in this area will lead to care that is better tailored to the individual patient, thereby making it more seamless and sustainable.

Third, many seams in our current care were identified. During interviews, the people with PD expressed a desire for more self management, better interdisciplinary collaboration between healthcare providers, more emphasis on prediction of disease development, and having access to a personal case manager. I believe that healthcare providers or organisations involved in PD should organize the care delivery in such way that such a dedicated care management professional can be created. Specialised PD nurses might be well suited to assume this role, but they might need to extend their focus to the social domain. Alternatively, rather than having a single case manager, it might be necessary to organise case management, whereby the responsibilities could be divided among for example a Parkinson nurse who would cover the medical domain in the hospital, and a general practitioner or a community nurse who covers the social and domestic domain. Regardless of the solution, introducing an optimal form of care management almost inevitably implies that care for people with PD should be moved more closely to the patient's home by introducing home-based care models. Technological innovations and self-monitoring apps can assist this important development.

Fourth, I think that the development or adoption of a broad OBPM might contribute to seamless care for people with PD. The current fee-for-service systems have important limitations regarding quality of care and healthcare costs. Broad OBPMs have proven to improve these limitations in other chronic conditions. I would recommend the development of a payment model that links financial incentives to quality indicators that are relevant for PD. These incentives should be combined with a global budget for PD care, with shared risks for providers and insurers.

Finally, I conclude that differences in density between individual provider networks for PD exist. Denser networks are associated with a lower risk of PD-related complications, higher healthcare utilization (of neurologists, physiotherapists and occupational therapists), and lower total healthcare costs for people with PD. Increasing density therefore has the potential to increase the value of care for individual people with PD. Selective purchasing methods aiming for higher density, could concentrate care among fewer providers. I suggest incorporating density as a quality indicator in an OBPM for PD.

Box 7.1: Suggestions for future research for seamless and sustainable care for people with PD

1. Examine the effects of possible sex-specific doctor' or patient' delays in PD to explain the 'earlier' use of multiple health services in women with PD.
2. Start a pilot in which specialized PD nurses act as a personal case manager for people with PD. Alternatively, perform experiments where case management for people with PD is ascertained, by dividing responsibilities across different domains.
3. Stimulate the use of technological innovations such as video consultations, wearable sensor technologies and self-monitoring apps, to further support the introduction of home-based care.
4. Design innovative care models that deliver care for people with PD more at home, with remote support by dedicated professional teams working either nearby in the community, a regional hospital or a remote university-based centre of expertise.
5. Develop an OBPM for PD care with density as an incorporated quality indicator, and study the effects on quality of care and healthcare costs.

If all of the above-mentioned suggestions are implemented, I have good hope that the care for people with PD will contain fewer and smaller seams than it has now, and that it will be more sustainable. I hope that future efforts of healthcare providers, policymakers, patient organizations, healthcare insurers, and – last but not least– people with PD and their caregivers will further contribute to the introduction of seamless and sustainable care for people with PD. Fortunately, I see great interest in developing and researching innovative care models. I also think that PD does not stand on itself. A better, more seamless and more sustainable care delivery for PD can serve as a blueprint for other chronic conditions, such as diabetes, COPD, or any chronic condition requiring a highly multidisciplinary approach.

Appendix

References

(CASP). 2017. 'CASP Qualitative Checklist', Critical Appraisal Skills Programme. http://www.casp-uk.net/casp-tools-checklists.

(CMS). 2016. 'Table 33: ACO Quality Measures', Centres for Medicare & Medicaid Services, Accessed 8-2-2017. https://www.cms.gov/Medicare/Medicare-Fee-for-Service-Payment/sharedsavingsprogram/Downloads/ACO-Shared-Savings-Program-Quality-Measures.pdf.

———. 2017. 'Hospital Value-Based Purchasing fact sheet', Centres for Medicare & Medicaid Services, Accessed 10-4-2018. https://www.cms.gov/Outreach-and-Education/Medicare-Learning-Network-MLN/MLNProducts/Downloads/Hospital_VBPurchasing_Fact_Sheet_ICN907664.pdf.

(HSCIC). 2014. "QOF indicator definitions 2014-15." In.: Health & Social Care Information Centre.

(OECD). 2011. "Country Classification 2011." In. Paris: Organisation for Economic Co-operation and Development.

———. 2014. "OECD Health Statistics 2014. Definitions, Sources and Methods Health Expenditure and Financing." In. Paris: Organisation for Economic Co-operation and Development.

Aarsland, D., K. Bronnick, J. P. Larsen, O. B. Tysnes, G. Alves, and Group Norwegian ParkWest Study. 2009. 'Cognitive impairment in incident, untreated Parkinson disease: the Norwegian ParkWest study', *Neurology*, 72: 1121-6.

Achey, M., J. L. Aldred, N. Aljehani, B. R. Bloem, K. M. Biglan, P. Chan, E. Cubo, E. R. Dorsey, C. G. Goetz, M. Guttman, A. Hassan, S. M. Khandhar, Z. Mari, M. Spindler, C. M. Tanner, P. van den Haak, R. Walker, and J. R. Wilkinson. 2014. 'The past, present, and future of telemedicine for Parkinson's disease', *Mov Disord*, 29: 871-83.

Addis, M. E., and J. R. Mahalik. 2003. 'Men, masculinity, and the contexts of help seeking', *Am Psychol*, 58: 5-14.

Afendulis, C. C., A. M. Fendrick, Z. Song, B. E. Landon, D. G. Safran, R. E. Mechanic, and M. E. Chernew. 2014. 'The impact of global budgets on pharmaceutical spending and utilization: early experience from the alternative quality contract', *Inquiry,* 51.

Alshamsan, R., J. T. Lee, A. Majeed, G. Netuveli, and C. Millett. 2012. 'Effect of a UK pay-for-performance program on ethnic disparities in diabetes outcomes: interrupted time series analysis', *Annals of Family Medicine,* 10: 228-34.

Alves, G., B. Müller, K. Herlofson, I. HogenEsch, W. Telstad, D. Aarsland, O.B. Tysnes, and J.P. Larsen. 2009. 'Incidence of Parkinson's disease in Norway: the Norwegian ParkWest study', *J Neurol Neurosurg Psychiatry.*, 80: 851-57.

Alzheimer Nederland. 2020. 'De casemanager: persoonlijke begeleiding bij dementie', "Alzheimer Nederland", Accessed 16-10. https://www.alzheimer-nederland.nl/dementie/diagnose-en-behandeling/casemanager.

Aminoff, M. J., C. W. Christine, J. H. Friedman, K. L. Chou, K. E. Lyons, R. Pahwa, B. R. Bloem, S. A. Parashos, C. C. Price, I. A. Malaty, R. Iansek, I. Bodis-Wollner, O. Suchowersky, W. H. Oertel, J. Zamudio, J. Oberdorf, P. Schmidt, M. S. Okun, and Disease National Parkinson Foundation Working Group on Hospitalization in Parkinson's. 2011. 'Management of the hospitalized patient with Parkinson's disease: current state of the field and need for guidelines', *Parkinsonism Relat Disord,* 17: 139-45.

Arora, S., V. Venkataraman, A. Zhan, S. Donohue, K. M. Biglan, E. R. Dorsey, and M. A. Little. 2015. 'Detecting and monitoring the symptoms of Parkinson's disease using smartphones: A pilot study', *Parkinsonism Relat Disord*, 21: 650-3.

Baba, Y., J.D. Putzke, N.R. Whaley, Z.K. Wszolek, and R.J. Uitti. 2005. 'Gender and the Parkinson's disease phenotype', *J Neurol.*, 252: 1201-05.

Baldereschi, Mm, A. Di Carlo, W.A. Rocca, P. Vanni, S. Maggi, E. Perissinotto, F. Grigoletto, L. Amaducci, and D. Inzitari. 2000. 'Parkinson's disease and parkinsonism in a longitudinal study: two-fold higher incidence in men. ILSA Working Group. Italian Longitudinal Study on Aging.', *Neurology,* 55: 1358-63.

Barnett, M. L., N. A. Christakis, J. O'Malley, J. P. Onnela, N. L. Keating, and B. E. Landon. 2012. 'Physician patient-sharing networks and the cost and intensity of care in US hospitals', *Med Care,* 50: 152-60.

Barnett, M. L., B. E. Landon, A. J. O'Malley, N. L. Keating, and N. A. Christakis. 2011. 'Mapping physician networks with self-reported and administrative data', *Health Serv Res,* 46: 1592-609.

Barry, C. L., E. A. Stuart, J. M. Donohue, S. F. Greenfield, E. Kouri, K. Duckworth, Z. Song, R. E. Mechanic, M. E. Chernew, and H. A. Huskamp. 2015. 'The Early Impact Of The 'Alternative Quality Contract' On Mental Health Service Use And Spending In Massachusetts', *Health Aff (Millwood),* 34: 2077-85.

Beersen, N., M. Berg, M. van Galen, K. Huijsmans, and N. Hoeksema. 2011. "Onderzoek naar de meerwaarde van ParkinsonNet." In, 1-42. Bilthoven, the Netherlands.

Bhattacharyya, T., P. Mehta, and A. A. Freiberg. 2008. 'Hospital characteristics associated with success in a pay-for-performance program in orthopaedic surgery', *Journal of Bone and Joint Surgery,* 90: 1240-43.

Bjornestad, Anders, Kenn Freddy Pedersen, Ole-Bjorn Tysnes, and Guido Alves. 2017. 'Clinical milestones in Parkinson's disease: A 7-year population-based incident cohort study', *Parkinsonism & Related Disorders,* 42: 28-33.

Bloem, B R., and F. Stocchi. 2012. 'Move for Change Part I: a European survey evaluating the impact of the EPDA Charter for People with Parkinson s disease', *European Journal of Neurology,* 19: 402-10.

———. 2015. 'Move for Change Part III: a European survey evaluating the impact of the EPDA Charter for People with Parkinson's Disease', *European Journal of Neurology,* 22: 133-41.

Bloem, B. R., N. M. de Vries, and G. Ebersbach. 2015. 'Nonpharmacological treatments for patients with Parkinson's disease', *Mov Disord,* 30: 1504-20.

Bloem, B. R., E. J. Henderson, E. R. Dorsey, M. S. Okun, N. Okubadejo, P. Chan, J. Andrejack, S. K. L. Darweesh, and M. Munneke. 2020. 'Integrated and patient-centred management of Parkinson's disease: a network model for reshaping chronic neurological care', *Lancet Neurol,* 19: 623-34.

Bloem, B. R., and M. Munneke. 2014. 'Revolutionising management of chronic disease: the ParkinsonNet approach', *BMJ,* 348: g1838.

Bloem, B. R., L. Rompen, N. M. Vries, A. Klink, M. Munneke, and P. Jeurissen. 2017. 'ParkinsonNet: A Low-Cost Health Care Innovation With A Systems Approach From The Netherlands', *Health Aff (Millwood),* 36: 1987-96.

Bloem, B. R., J. H. L. Ypinga, A. Willis, C. G. Canning, R. A. Barker, M. Munneke, and N. M. De Vries. 2018. 'Using Medical Claims Analyses to Understand Interventions for Parkinson Patients', *J Parkinsons Dis,* 8: 45-58.

Bloem, B.R., T. van Laar, S.H.J. Keus, H. de Beer, E. Poot, E. Buskens, W. Aarden, and M. Munneke. 2010. "Multidisciplinaire richtlijn 'Ziekte van Parkinson'." In. Alphen aan den Rijn.

Buetow, S., L. S. Giddings, L. Williams, and S. Nayar. 2008. 'Perceived unmet needs for health care among Parkinson's Society of New Zealand members with Parkinson's disease', *Parkinsonism Relat Disord,* 14: 495-500.

Calikoglu, S., R. Murray, and D. Feeney. 2012. 'Hospital pay-for-performance programs in Maryland produced strong results, including reduced Hospital-Acquired conditions', *Health affairs,* 31: 2649-58.

Campbell, S. M., R. McDonald, and H. Lester. 2008. 'The experience of pay for performance in english family practice: A qualitative study', *Annals of Family Medicine,* 6: 228-34.

Campbell, S. M., D. Reeves, E. Kontopantelis, B. Sibbald, and M. Roland. 2009. 'Effects of pay for performance on the quality of primary care in England', *N Engl J Med,* 361: 368-78.

Canoy, M., M. J. Faber, M. Munneke, W. Oortwijn, M. J. Nijkrake, and B. R. Bloem. 2015. 'Hidden Treasures and Secret Pitfalls: Application of the Capability Approach to ParkinsonNet', *J Parkinsons Dis,* 5: 575-80.

CBS. 2018. 'Bevolkingsteller', Centraal Bureau voor de Statistiek, Accessed 10-01-2019. https://www.cbs.nl/nl-nl/visualisaties/bevolkingsteller.

———. 2020. 'Bevolkingsteller', Centraal Bureau voor de Statistiek, Accessed 10-08-2020. https://www.cbs.nl/nl-nl/visualisaties/bevolkingsteller.

Chatfield, J. S. 2016. 'Value-Based Purchasing: The Effect of Hospital Ownership and Size', *Health Care Manag.,* 35: 199-205.

Chee, T. T., A. M. Ryan, J. H. Wasfy, and W. B. Borden. 2016. 'Current State of Value-Based Purchasing Programs', *Circulation,* 133: 2197-205.

Chien, A. T., D. Eastman, Z. Li, and M. B. Rosenthal. 2012. 'Impact of a pay for performance program to improve diabetes care in the safety net', *Preventive medicine,* 55: S80-S85.

Chien, A. T., Z. Li, and M. B. Rosenthal. 2010. 'Improving timely childhood immunizations through pay for performance in medicaid-managed care', *Health Services Research,* 45: 1934-47.

Chien, A. T., K. H. Schiavoni, E. Sprecher, B. E. Landon, B. J. McNeil, M. E. Chernew, and M. A. Schuster. 2016. 'How Accountable Care Organizations Responded to Pediatric Incentives in the Alternative Quality Contract', *Acad Pediatr,* 16: 200-7.

Chien, A. T., Z. Song, M. E. Chernew, B. E. Landon, B. J. McNeil, D. G. Safran, and M. A. Schuster. 2014. 'Two-year impact of the alternative quality contract on pediatric health care quality and spending', *Pediatrics,* 133: 96-104.

Chou, K.L., H.I. Hurtig, and A.F. Eichler. 2019. "Clinical manifestations of Parkinson disease." In. Alphen aan den Rijn: UpToDate.

Chung, S., L. P. Palaniappan, L. M. Trujillo, H. R. Rubin, and H. S. Luft. 2010. 'Effect of physician-specific pay-for-performance incentives in a large group practice', *The American journal of managed care,* 16: 35-42.

Chung, S., L. Palaniappan, E. Wong, H. Rubin, and H. Luft. 2010. 'Does the frequency of pay-for-performance payment matter? Experience from a randomized trial', *Health Services Research,* 45: 553-64.

Cohen, D. 2009. 'Men need primary care at work, debate hears', *BMJ,* 338: b2471.

Colombo, Francesca, and Organisation for Economic Co-operation and Development. 2011. *Help wanted? : providing and paying for long-term care* (OECD: Paris).

Conrad, D. A. , and L. Perry. 2009. 'Quality-based financial incentives in health care: can we improve quality by paying for it?', *Annu Rev Public Health.,* 30: 357-71.

Constantinescu, G., D. Theodoros, T. Russell, E. Ward, S. Wilson, and R. Wootton. 2011. 'Treating disordered speech and voice in Parkinson's disease online: a randomized controlled non-inferiority trial', *Int J Lang Commun Disord,* 46: 1-16.

Crawley, D., A. Ng, A. G. Mainous, A. Majeed, and C. Millett. 2009. 'Impact of pay for performance on quality of chronic disease management by social class group in England', *Journal of the Royal Society of Medicine,* 102: 103-07.

Crepaz, M., and J. M. Curry. 2013. 'Improving the cancer patient journey', *Stud Health Technol Inform,* 188: 14-9.

Crizzle, A. M., S. Classen, and E. Y. Uc. 2012. 'Parkinson disease and driving: an evidence-based review', *Neurology,* 79: 2067-74.

Cryer, L., S. B. Shannon, M. Van Amsterdam, and B. Leff. 2012. 'Costs for 'hospital at home' patients were 19 percent lower, with equal or better outcomes compared to similar inpatients', *Health Aff (Millwood),* 31: 1237-43.

Cubo, Esther. 'Task Force on Telemedicine', International Parkinson and Movement Disorder Society Accessed Jun 9. http://www.movementdisorders.org/MDS/About/Committees--Other-Groups/MDS-Task-Forces/Task-Force-on-Telemedicine.htm.

Darkins, A., P. Ryan, R. Kobb, L. Foster, E. Edmonson, B. Wakefield, and A. E. Lancaster. 2008. 'Care Coordination/Home Telehealth: the systematic implementation of health informatics, home telehealth, and disease management to support the care of veteran patients with chronic conditions', *Telemed J E Health,* 14: 1118-26.

Darweesh, S. K., V. J. Verlinden, B. H. Stricker, A. Hofman, P. J. Koudstaal, and M. A. Ikram. 2017. 'Trajectories of prediagnostic functioning in Parkinson's disease', *Brain,* 140: 429-41.

de Lau, L. M., and M. M. Breteler. 2006. 'Epidemiology of Parkinson's disease', *Lancet Neurol,* 5: 525-35.

de Lau, L. M., C. M. Schipper, A. Hofman, P. J. Koudstaal, and M. M. Breteler. 2005. 'Prognosis of Parkinson disease: risk of dementia and mortality: the Rotterdam Study', *Arch Neurol,* 62: 1265-9.

Donabedian, A. 1988. 'The quality of care: How can it be assessed?', *JAMA.,* 260: 1743–8.

Doran, T., C. Fullwood, H. Gravelle, D. Reeves, E. Kontopantelis, U. Hiroeh, and M. Roland. 2006. 'Pay-for-performance programs in family practices in the United Kingdom', *New England Journal of Medicine,* 355: 375-84.

Doran, T., C. Fullwood, E. Kontopantelis, and D. Reeves. 2008. 'Effect of financial incentives on inequalities in the delivery of primary clinical care in England: analysis of clinical activity indicators for the quality and outcomes framework', *Lancet,* 372: 728-36.

Doran, T., C. Fullwood, D. Reeves, H. Gravelle, and M. Roland. 2008. 'Exclusion of patients from pay-for-performance targets by english physicians', *New England Journal of Medicine,* 359: 274-84.

Doran, T., E. Kontopantelis, C. Fullwood, H. Lester, J. M. Valderas, and S. Campbell. 2012. 'Exempting dissenting patients from pay for performance schemes: retrospective analysis of exception reporting in the UK Quality and Outcomes Framework', *BMJ (Clinical research ed.),* 344: e2405.

Doran, T., E. Kontopantelis, J. M. Valderas, S. Campbell, M. Roland, C. Salisbury, and D. Reeves. 2011. 'Effect of financial incentives on incentivised and non-incentivised clinical activities: Longitudinal analysis of data from the UK Quality and Outcomes Framework', *BMJ,* 343.

Dorsey, E. R., M. A. Achey, C. A. Beck, D. B. Beran, K. M. Biglan, C. M. Boyd, P. N. Schmidt, R. Simone, A. W. Willis, N. B. Galifianakis, M. Katz, C. M. Tanner, K. Dodenhoff, N. Ziman, J. Aldred, J. Carter, J. Jimenez-Shahed, C. Hunter, M. Spindler, Z. Mari, J. C. Morgan, D. McLane, P. Hickey, L. Gauger, I. H. Richard, M. T. Bull, N. I. Mejia, G. Bwala, M. Nance, L. Shih, L. Anderson, C. Singer, C. Zadikoff, N. Okon, A. Feigin, J. Ayan, C. Vaughan, R. Pahwa, J. Cooper, S. Webb, R. Dhall, A. Hassan, D. Weis, S. DeMello, S. S. Riggare, P. Wicks, J. Smith, H. T. Keenan, R. Korn, H. Schwarz, S. Sharma, E. A. Stevenson, and W. Zhu. 2016. 'National Randomized Controlled Trial of Virtual House Calls for People with Parkinson's Disease: Interest and Barriers', *Telemed J E Health*.

Dorsey, E. R., R. Constantinescu, J. P. Thompson, K. M. Biglan, R. G. Holloway, K. Kieburtz, F. J. Marshall, B. M. Ravina, G. Schifitto, A. Siderowf, and C. M. Tanner. 2007. 'Projected number of people with Parkinson disease in the most populous nations, 2005 through 2030', *Neurology,* 68: 384-6.

Dorsey, E. R., L. M. Deuel, T. S. Voss, K. Finnigan, B. P. George, S. Eason, D. Miller, J. I. Reminick, A. Appler, J. Polanowicz, L. Viti, S. Smith, A. Joseph, and K. M. Biglan. 2010. 'Increasing access to specialty care: a pilot, randomized controlled trial of telemedicine for Parkinson's disease', *Mov Disord,* 25: 1652-9.

Dorsey, E. R., T. Sherer, M. S. Okun, and B. R. Bloem. 2018. 'The Emerging Evidence of the Parkinson Pandemic', *J Parkinsons Dis,* 8: S3-s8.

Dorsey, E. R., V. Venkataraman, M. J. Grana, M. T. Bull, B. P. George, C. M. Boyd, C. A. Beck, B. Rajan, A. Seidmann, and K. M. Biglan. 2013. 'Randomized controlled clinical trial of "virtual house calls" for Parkinson disease', *JAMA Neurol,* 70: 565-70.

Dorsey, E. R., and A. W. Willis. 2013. 'Caring for the majority', Mov Disord, 28: 261-2.

Dorsey, E. Ray, Floris P. Vlaanderen, Lucien Jlpg Engelen, Karl Kieburtz, William Zhu, Kevin M. Biglan, Marjan J. Faber, and Bastiaan R. Bloem. 2016. 'Moving Parkinson care to the home', *Movement disorders : official journal of the Movement Disorder Society,* 31: 1258-62.

Dorsey, E.R., T. Sherer, M.S. Okun, and B.R. Boem. 2020. *Ending Parkinson's disease* (Ingram Publisher Services US Jackson, USA).

Downs, S. H., and N. Black. 1998. 'The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions', *J Epidemiol Community Health,* 52: 377–84.

Eggers, C., R. Dano, J. Schill, G. R. Fink, M. Hellmich, and L. Timmermann. 2018. 'Patient-centered integrated healthcare improves quality of life in Parkinson's disease patients: a randomized controlled trial', *J Neurol,* 265: 764-73.

Eijkenaar, F. 2012. 'Pay for performance in health care: an international overview of initiatives', *Med Care Res Rev.*, 69: 251-76.

———. 2013. 'Key issues in the design of pay for performance programs.', *Eur J Health Econ.*, Feb 14: 117-31.

Eijkenaar, F., M. Emmert, M. Scheppach, and O. Schoffski. 2013. 'Effects of pay for performance in health care: a systematic review of systematic reviews', *Health Policy,* 110: 115-30.

Eijkenaar, F., and F. T. Schut. 2015. "Uitkomstbekostiging in de zorg: een (on)begaanbare weg?" In. Rotterdam: instituut Beleid & Management in de Gezondheidszorg.

Eijkenaar, F., W. van de Ven, and F. T. Schut. 2012. "Uitkomstbekostiging in de zorg: internationale voorbeelden en relevantie voor Nederland." In. Rotterdam: instituut Beleid & Management in de Gezondheidszorg.

Epstein, A. M., A. K. Jha, and E. J. Orav. 2014. 'The impact of pay-for-performance on quality of care for minority patients', *Am J Manag Care,* 20: 479-86.

Epstein, A. M., K. E. Joynt, A. K. Jha, and E. J. Orav. 2014. 'Access to coronary artery bypass graft surgery under pay for performance: evidence from the premier hospital quality incentive demonstration', *Circ Cardiovasc Qual Outcomes,* 7: 727-34.

Espay, A. J., P. Bonato, F. B. Nahab, W. Maetzler, J. M. Dean, J. Klucken, B. M. Eskofier, A. Merola, F. Horak, A. E. Lang, R. Reilmann, J. Giuffrida, A. Nieuwboer, M. Horne, M. A. Little, I. Litvan, T. Simuni, E. R. Dorsey, M. A. Burack, K. Kubota, A. Kamondi, C. Godinho, J. F. Daneault, G. Mitsi, L. Krinke, J. M. Hausdorff, B. R. Bloem, S. Papapetropoulos, and Technology Movement Disorders Society Task Force on. 2016. 'Technology in Parkinson's disease: Challenges and opportunities', *Mov Disord*.

'The European Parkinson's disease Standards of Care Consensus Statement'. 2011. European Parkinson's Disease Association, Accessed June 6. http://alt.kompetenznetz-parkinson.de/EPDA_Parkinson_s_Standard_nsus_Statement_Vol_I.pdf.

Ferreira, J. J., C. Godinho, A. T. Santos, J. Domingos, D. Abreu, R. Lobo, N. Goncalves, M. Barra, F. Larsen, O. Fagerbakke, I. Akeren, H. Wangen, J. A. Serrano, P. Weber, A. Thoms, S. Meckler, S. Sollinger, J. van Uem, M. A. Hobert, K. S. Maier, H. Matthew, T. Isaacs, J. Duffen, H. Graessner, and W. Maetzler. 2015. 'Quantitative home-based assessment of Parkinson's symptoms: the SENSE-PARK feasibility and usability study', *BMC Neurol,* 15: 89.

Figueroa, J. F., Y. Tsugawa, J. Zheng, E. J. Orav, and A. K. Jha. 2016. 'Association between the Value-Based Purchasing pay for performance program and patient mortality in US hospitals: observational study', *BMJ,* 353.

Findley, L. J., and M. G. Baker. 2002. 'Treating neurodegenerative diseases', *BMJ,* 324: 1466-7.

Fleetcroft, R., N. Steel, R. Cookson, S. Walker, and A. Howe. 2012. 'Incentive payments are not related to expected health gain in the pay for performance scheme for UK primary care: cross-sectional analysis', *BMC Health Services Research,* 12: 94.

Fleisher, J., W. Barbosa, M. M. Sweeney, S. E. Oyler, A. C. Lemen, A. Fazl, M. Ko, T. Meisel, N. Friede, G. Dacpano, R. M. Gilbert, A. Di Rocco, and J. Chodosh. 2018. 'Interdisciplinary Home Visits for Individuals with Advanced Parkinson's Disease and Related Disorders', *J Am Geriatr Soc,* 66: 1226-32.

Fleisher, J., D. Tarsy, H.I. Hurtig, and A.F. Eichler. 2019. 'Nonpharmacologic management of Parkinson disease', UpToDate, Accessed 06 march.

Fox, S., and K. Purcell. 2010. 'Chronic Disease and the Internet ', Pew Research Center: Internet, Science & Tech, Accessed Jun 6. http://www.pewinternet.org/files/old-media/Files/Reports/2010/PIP_Chronic_Disease_with_topline.pdf.

Foy, R., S. Hempel, L. Rubenstein, M. Suttorp, M. Seelig, R. Shanman, and P. G. Shekelle. 2010. 'Meta-analysis: effect of interactive communication between collaborating primary care physicians and specialists', *Ann Intern Med,* 152: 247-58.

Fullard, M. E., D. P. Thibault, A. Hill, J. Fox, D. E. Bhatti, M. A. Burack, N. Dahodwala, E. Haberfeld, D. S. Kern, O. S. Klepitskava, E. Urrea-Mendoza, P. Myers, J. Nutt, M. R. Rafferty, J. M. Schwalb, L. M. Shulman, and A. W. Willis. 2017. 'Utilization of rehabilitation therapy services in Parkinson disease in the United States', *Neurology,* 89: 1162-69.

Gal, O., M. Srp, R. Konvalinkova, M. Hoskovcova, V. Capek, J. Roth, and E. Ruzicka. 2017. 'Physiotherapy in Parkinson's Disease: Building ParkinsonNet in Czechia', *Parkinsons Dis,* 2017: 8921932.

Gallagher, N., C. Cardwell, C. Hughes, and D. O'Reilly. 2014. 'Increase in the pharmacological management of Type 2 diabetes with pay-for-performance in primary care in the UK', *Diabetic Medicine,* 32: 62-68.

Georgiev, D., K. Hamberg, M. Hariz, L. Forsgren, and Hariz G-M. 2017. 'Gender differences in Parkinson's disease: A clinical perspective', *Acta Neurol Scand,* 136: 570–84.

Gerlach, O. H., A. Winogrodzka, and W. E. Weber. 2011. 'Clinical problems in the hospitalized Parkinson's disease patient: systematic review', *Mov Disord,* 26: 197-208.

Gillam, S. J., A. N. Siriwardena, and N. Steel. 2012. 'Pay-for-performance in the United Kingdom: impact of the quality and outcomes framework: a systematic review', *Annals of Family Medicine,* 10: 461-68.

Gilman, M., E. K. Adams, J. M. Hockenberry, A. S. Milstein, I. B. Wilson, and E. R. Becker. 2015. 'Safety-net hospitals more likely than other hospitals to fare poorly under medicare's value-based purchasing', *Health Affairs,* 34: 398-405.

Göttgens, I., A. D. van Halteren, N. M. de Vries, M. J. Meinders, Y. Ben-Shlomo, B. R. Bloem, S. K. L. Darweesh, and S. Oertelt-Prigione. 2020. 'The Impact of Sex and Gender on the Multidisciplinary Management of Care for Persons With Parkinson's Disease', *Front Neurol,* 11: 576121.

Gravelle, Hugh, Matt Sutton, and Ada Ma. 2010. 'Doctor Behaviour under a Pay for Performance Contract: Treating, Cheating and Case Finding?', *Economic Journal,* 120: F129-56.

Greene, J., J. H. Hibbard, and V. Overton. 2015. 'Large performance incentives had the greatest impact on providers whose quality metrics were lowest at baseline', *Health Aff.,* 34: 673-80.

Grosset, K. A., and D. G. Grosset. 2005. 'Patient-perceived involvement and satisfaction in Parkinson's disease: effect on therapy decisions and quality of life', *Mov Disord,* 20: 616-9.

Haaxma, C.A., B.R. Bloem, G.F. Borm, W.J. Oyen, K.L. Leenders, S. Eshuis, J. Booij, D.E. Dluzen, and M.W. Horstink. 2007. 'Gender differences in Parkinson's disease.', *J Neurol Neurosurg Psychiatry.,* 78: 819-24.

Hack, N., U. Akbar, E. H. Monari, A. Eilers, A. Thompson-Avila, N. H. Hwynn, A. Sriram, I. Haq, A. Hardwick, I. A. Malaty, and M. S. Okun. 2015. 'Person-Centered Care in the Home Setting for Parkinson's Disease: Operation House Call Quality of Care Pilot Study', *Parkinsons Dis,* 2015: 639494.

Hammond, W. E. 2010. 'Seamless care: what is it; what is its value; what does it require; when might we get it?', *Stud Health Technol Inform,* 155: 3-13.

Harrison, M. J., M. Dusheiko, M. Sutton, H. Gravelle, T. Doran, and M. Roland. 2014. 'Effect of a national primary care pay for performance scheme on emergency hospital admissions for ambulatory care sensitive conditions: controlled longitudinal study', *BMJ,* 349.

Hayen, A. P., P. J. G. M. de Bekker, M. M. T. J. Ouwens, G. P. Westert, and P. P. T. Jeurissen. 2013. "No cure, no pay? The road to outcome-based payments in Dutch healthcare; current situation and opportunities." In. Nijmegen: Cesus academy for sustainable care.

"Health Care's Big Spenders: The Characteristics Behind the Curve." In. 2016.

Hibbard, J. H., E. Mahoney, and E. Sonet. 2017. 'Does patient activation level affect the cancer patient journey?', *Patient Educ Couns,* 100: 1276-79.

Hijdra, A., P.J. Koudstaal, and R.A.C. Roos. 2012. *Neurologie* (Elsevier Gezondheidszorg: Amsterdam).

———. 2016. *Neurologie* (Elsevier Gezondheidszorg: Amsterdam).

Howitt, M. J. 2011. 'The family care coordinator: paving the way to seamless care', *J Pediatr Oncol Nurs,* 28: 107-13.

Huber, Machteld, J André Knottnerus, Lawrence Green, Henriëtte van der Horst, Alejandro R Jadad, Daan Kromhout, Brian Leonard, Kate Lorig, Maria Isabel Loureiro, Jos W M van der Meer, Paul Schnabel, Richard Smith, Chris van Weel, and Henk Smid. 2011. 'How should we define health?', *BMJ,* 343.

Institute of Medicine (U.S.). Committee on Quality of Health Care in America. 2001. *Crossing the quality chasm : a new health system for the 21st century* (National Academy Press: Washington, D.C.).

Jankovic, J., H.I. Hurtig, and A.F. Eichler. 2019. 'Etiology and pathogenesis of Parkinson disease', UpToDate, Accessed 10 august.

Jeurissen, P., H. Maarse, and M. Tanke. 2018. *Betaalbare zorg* (Sdu: The Hague, the Netherlands).

Jha, A. K. 2013. 'Time to get serious about pay for performance.', *JAMA,* 309: 347-8.

Jha, A. K., K. E. Joynt, E. J. Orav, and A. M. Epstein. 2012. 'The long-term effect of premier pay for performance on patient outcomes', *New England Journal of Medicine,* 366: 1606-15.

Jolly, Jennifer. 2015. "An Uber for Doctor Housecalls." In *Wired Well*. The New York Times The New York Times Company.

Kahn, C. N., T. Ault, L. Potetz, T. Walke, J. H. Chambers, and S. Burch. 2015. 'Assessing Medicare's hospital pay-for-performance programs and whether they are achieving their goals', *Health Aff.,* 34: 1281-8.

Kaiser Permanente. 2016. 'Kaiser Permanente moms and families help design care at Baldwin Park Medical Center', Accessed April 30, 2018. https://share.kaiserpermanente.org/article/kaiser-permanente-moms-and-families-help-design-care-at-baldwin-park-medical-center/.

Kalf, J.G., B.J.M. de Swart, M. Bonnier, M. Hofman, J. Kanters, J. Kocken, M. Miltenburg, B.R. Bloem, and M. Munneke. 2008. "Guidelines for Speech-Language Therapy in Parkinson's Disease." In. Nijmegen, The Netherlands / Miami (FL), U.S.A.: ParkinsonNet/NPF.

Karunaratne, K., P. Stevens, J. Irving, H. Hobbs, H. Kilbride, R. Kingston, and C. Farmer. 2013. 'The impact of pay for performance on the control of blood pressure in people with chronic kidney disease stage 3-5', *Nephrology Dialysis Transplantation,* 28: 2107-16.

Kasteridis, P., A. Mason, M. Goddard, R. Jacobs, R. Santos, B. Rodriguez-Sanchez, and G. McGonigal. 2016. 'Risk of Care Home Placement following Acute Hospital Admission: Effects of a Pay-for-Performance Scheme for Dementia', *PLoS One,* 11.

Kelly, J., J. Dwyer, T. Mackean, O. Donnell K, and E. Willis. 2017. 'Coproducing Aboriginal patient journey mapping tools for improved quality and coordination of care', *Aust J Prim Health,* 23: 536-42.

Kent, Lauren. 2015. 'Where do the oldest Americans live?', Pew Research Center, Accessed Jun 9. http://www.pewresearch.org/fact-tank/2015/07/09/where-do-the-oldest-americans-live/.

Ketelaar, N. A., M. Munneke, B. R. Bloem, G. P. Westert, and M. J. Faber. 2013. 'Recognition of physiotherapists' expertise in Parkinson's disease', *BMC Health Serv Res,* 13: 430.

Keus, S. H., B. R. Bloem, D. Verbaan, P. A. de Jonge, M. Hofman, B. J. van Hilten, and M. Munneke. 2004. 'Physiotherapy in Parkinson's disease: utilisation and patient satisfaction', *J Neurol,* 251: 680-7.

Keus, S.H.J., M. Munneke, M. Graziano, J. Paltamaa, E. Pelosin, J. Domingos, S. Brühlmann, B. Ramaswamy, J. Prins, C. Struiksma, L. Rochester, A. Nieuwboer, and B.R. Bloem. 2014. "European Physiotherapy Guideline for Parkinson's Disease." In. the Netherlands: *KNGF/ParkinsonNet*.

Kim, S. M., W. M. Jang, H. A. Ahn, H. J. Park, and H. S. Ahn. 2012. 'Korean National Health Insurance Value Incentive Program: Achievements and future directions', *Journal of Preventive Medicine and Public Health,* 45: 148-55.

Kleiner-Fisman, G., P. Gryfe, and G. Naglie. 2013. 'A Patient-Based Needs Assessment for Living Well with Parkinson Disease: Implementation via Nominal Group Technique', *Parkinsons Dis,* 2013: 974964.

Knight, D. A., D. Thompson, E. Mathie, and A. Dickinson. 2013. "Seamless care? Just a list would have helped!' Older people and their carer's experiences of support with medication on discharge home from hospital', *Health Expect,* 16: 277-91.

Kovács, M., A. Makkos, Z. Aschermann, J. Janszky, S. Komoly, R. Weintraut, K. Karádi, and N. Kovács. 2016. 'Impact of Sex on the Nonmotor Symptoms and the Health-Related Quality of Life in Parkinson's Disease.', *Parkinsons Dis,* 2016: 7951840.

Kowal, S. L., T. M. Dall, R. Chakrabarti, M. V. Storm, and A. Jain. 2013. 'The current and projected economic burden of Parkinson's disease in the United States', *Mov Disord,* 28: 311-8.

Kristensen, S. R., R. McDonald, and M. Sutton. 2013. 'Should pay-for-performance schemes be locally designed? Evidence from the Commissioning for Quality and Innovation (CQUIN) Framework', *Journal of health services research & policy,* 18: 38-49.

Kruse, G. B., D. Polsky, E. A. Stuart, and R. M. Werner. 2012. 'The Impact of Hospital Pay-for-Performance on Hospital and Medicare Costs', *Health Services Research,* 47: 2118-36.

Kuo, S., K. E. Huang, S. A. Davis, and S. R. Feldman. 2015. 'The rosacea patient journey: a novel approach to conceptualizing patient experiences', *Cutis,* 95: 37-43.

Landers, Steven H. 2010. 'Why Health Care Is Going Home', *New England Journal of Medicine,* 363: 1690-91.

Landers, Steven, Elizabeth Madigan, Bruce Leff, Robert J. Rosati, Barbara A. McCann, Rodney Hornbake, Richard MacMillan, Kate Jones, Kathryn Bowles, Dawn Dowding, Teresa Lee, Tracey Moorhead, Sally Rodriguez, and Erica Breese. 2016. 'The Future of Home Health Care: A Strategic Framework for Optimizing Value', *Home health care management & practice,* 28: 262-78.

Landon, B. E., N. L. Keating, M. L. Barnett, J. P. Onnela, S. Paul, A. J. O'Malley, T. Keegan, and N. A. Christakis. 2012. 'Variation in patient-sharing networks of physicians across the United States', *JAMA,* 308: 265-73.

Landon, B. E., N. L. Keating, J. P. Onnela, A. M. Zaslavsky, N. A. Christakis, and A. J. O'Malley. 2018. 'Patient-Sharing Networks of Physicians and Health Care Utilization and Spending Among Medicare Beneficiaries', *JAMA Intern Med,* 178: 66-73.

Laveau, F., N. Hammoudi, E. Berthelot, J. Belmin, P. Assayag, A. Cohen, T. Damy, D. Duboc, O. Dubourg, A. Hagege, O. Hanon, R. Isnard, G. Jondeau, F. Labouree, D. Logeart, N. Mansencal, C. Meune, E. Pautas, Y. Wolmark, and M. Komajda. 2017. 'Patient journey in decompensated heart failure: An analysis in departments of cardiology and geriatrics in the Greater Paris University Hospitals', *Arch Cardiovasc Dis,* 110: 42-50.

Lee, J. T., G. Netuveli, A. Majeed, and C. Millett. 2011. 'The effects of pay for performance on disparities in stroke, hypertension, and coronary heart disease management: Interrupted time series study', *PloS one,* 6.

Lee, J. Y., S. I. Lee, N. S. Kim, S. H. Kim, W. S. Son, and M. W. Jo. 2012. 'Healthcare organizations' attitudes toward pay-for-performance in Korea', *Health Policy,* 108: 277-85.

Lees, A. J., J. Hardy, and T. Revesz. 2009. 'Parkinson's disease', Lancet, 373: 2055-66.

Levitz, J. . "Communities Struggle to Care for Elderly, Alone at Home: More people age at home, raising demand for support services." In.: The Wall Street Journal.

Maarse, H., P. Jeurissen, and D. Ruwaard. 2016. 'Results of the market-oriented reform in the Netherlands: a review', *Health Econ Policy Law,* 11: 161-78.

MacNeil Vroomen, Janet, Judith E. Bosmans, Iris Eekhout, Karlijn J. Joling, Lisa D. van Mierlo, Franka J. M. Meiland, Hein P. J. van Hout, and Sophia E. de Rooij. 2016. 'The Cost-Effectiveness of Two Forms of Case Management Compared to a Control Group for Persons with Dementia and Their Informal Caregivers from a Societal Perspective', *PLOS ONE*, 11: e0160908.

Maetzler, W., J. Domingos, K. Srulijes, J. J. Ferreira, and B. R. Bloem. 2013. 'Quantitative wearable sensors for objective assessment of Parkinson's disease', *Mov Disord,* 28: 1628-37.

Mancini, F., A. D. van Halteren, T. Carta, S. Thomas, B. R. Bloem, and S. K. L. Darweesh. 2020. 'Personalized care management for persons with Parkinson's disease: A telenursing solution', *Clin Park Relat Disord,* 3: 100070.

Mant, J. 2001. 'Process versus outcome indicators in the assessment of quality of health care', *International Journal for Quality in Health Care,* 13.

Marras, C., J.C. Beck, J.H. Bower, E. Roberts, B. Ritz, G.W. Ross, R.D. Abbott, R. Savica, S.K. Van Den Eeden, A.W. Willis, and C.M. Tanner. 2018. 'Prevalence of Parkinson's disease across North America', *NPJ Parkinsons Dis.,* 4: 21.

Martinez-Martin, P., M. J. Forjaz, B. Frades-Payo, A. B. Rusinol, J. M. Fernandez-Garcia, J. Benito-Leon, V. C. Arillo, M. A. Barbera, M. P. Sordo, and M. J. Catalan. 2007. 'Caregiver burden in Parkinson's disease', *Mov Disord,* 22: 924-31; quiz 1060.

McCarthy, M. 2015. '"Value based" payment project saves US nearly $400m in two years, report finds', *BMJ,* 350: h2432.

McDonald, R., S. R. Kristensen, S. Zaidi, M. Sutton, S. Todd, F. Konteh, K. Hussein, and S. Brown. 2013. "Evaluation of the Commissioning for Quality and Innovation Framework." In.

McDonald, R., and M. Roland. 2009. 'Pay for performance in primary care in England and california: Comparison of unintended consequences', *Annals of Family Medicine,* 7: 121-27.

McWilliams, J. M., B. E. Landon, and M. E. Chernew. 2013. 'Changes in health care spending and quality for medicare beneficiaries associated with a commercial ACO contract', *JAMA,* 310: 829-36.

Mehrotra, A., C. L. Damberg, M. E. S. Sorbero, and S. S. Teleki. 2009. 'Pay for performance in the hospital setting: What is the state of the evidence?', *American Journal of Medical Quality,* 24: 19-28.

Mehrotra, A., M. E. Sorbero, and C. L. Damberg. 2010. 'Using the lessons of behavioral economics to design more effective pay-for-performance programs', *Am J Manag Care.,* 16: 497-503.

Mehta, A., N. Belmatoug, B. Bembi, P. Deegan, D. Elstein, O. Goker-Alpan, E. Lukina, E. Mengel, K. Nakamura, G. M. Pastores, J. Perez-Lopez, I. Schwartz, C. Serratrice, J. Szer, A. Zimran, M. Di Rocco, Z. Panahloo, D. J. Kuter, and D. Hughes. 2017. 'Exploring the patient journey to diagnosis of Gaucher disease from the perspective of 212 patients with Gaucher disease and 16 Gaucher expert physicians', *Mol Genet Metab,* 122: 122-29.

Mellor, J., M. Daly, and M. Smith. 2016. 'Does It Pay to Penalize Hospitals for Excess Readmissions? Intended and Unintended Consequences of Medicare's Hospital Readmissions Reductions Program', *Health Econ,* Jul 15.

Meyer, G. S., and R. V. Gibbons. 1997. 'House calls to the elderly--a vanishing practice among physicians', *N Engl J Med,* 337: 1815-20.

Millett, C., A. Bottle, A. Ng, V. Curcin, M. Molokhia, S. Saxena, and A. Majeed. 2009. 'Pay for perfomance and the quality of diabetes management in individuals with and without co-morbid medical conditions', *Journal of the Royal Society of Medicine,* 102: 369-77.

Millett, C., J. Gray, A. Bottle, and A. Majeed. 2008. 'Ethnic disparities in blood pressure management in patients with hypertension after the introduction of pay for performance', *Annals of Family Medicine,* 6: 490-96.

Millett, C., J. Gray, S. Saxena, G. Netuveli, K. Khunti, and A. Majeed. 2007. 'Ethnic disparities in diabetes management and pay-for-performance in the UK: the Wandsworth Prospective Diabetes Study', *PLoS medicine,* 4.

Millett, C., J. Gray, S. Saxena, G. Netuveli, and A. Majeed. 2007. 'Impact of a pay-for-performance incentive on support for smoking cessation and on smoking prevalence among people with diabetes', *Cmaj,* 176: 1705-10.

Millett, C., S. Saxena, G. Netuveli, and A. Majeed. 2009. 'Impact of pay for performance on ethnic disparities in intermediate outcomes for diabetes: A longitudinal study', *Diabetes Care,* 32: 404-09.

Milstein, R.;, and J. Schreyoegg. 2016. 'Pay for performance in the inpatient sector: A review of 34 P4P programs in 14 OECD countries', *Health Policy,* 120: 1125–40.

Miyasaki, J. M., J. Long, D. Mancini, E. Moro, S. H. Fox, A. E. Lang, C. Marras, R. Chen, A. Strafella, R. Arshinoff, R. Ghoche, and J. Hui. 2012. 'Palliative care for advanced Parkinson disease: an interdisciplinary clinic and new scale, the ESAS-PD', *Parkinsonism Relat Disord,* 18 Suppl 3: S6-9.

Moen, E. L., and J. P. W. Bynum. 2019. 'Evaluation of Physician Network-Based Measures of Care Coordination Using Medicare Patient-Reported Experience Measures', *J Gen Intern Med,* 34: 2482-89.

Mosca, I., P. J. van der Wees, E. S. Mot, J. J. G. Wammes, and P. P. T. Jeurissen. 2016. 'Sustainability of Long-term Care: Puzzling Tasks Ahead for Policy-Makers', *Int J Health Policy Manag,* 6: 195-205.

Munneke, M., M. J. Nijkrake, S. H. Keus, G. Kwakkel, H. W. Berendse, R. A. Roos, G. F. Borm, E. M. Adang, S. Overeem, and B. R. Bloem. 2010. 'Efficacy of community-based physiotherapy networks for patients with Parkinson's disease: a cluster-randomised trial', *Lancet Neurol,* 9: 46-54.

Muzerengi, S., C. Herd, C. Rick, and C. E. Clarke. 2016. 'A systematic review of interventions to reduce hospitalisation in Parkinson's disease', P*arkinsonism Relat Disord,* 24: 3-7.

National Institute of Public Health and the Environment. 2019a. "Cost of illness tool 2017." In. Bilthoven.

———. 2019b. 'Kosten van ziekten | Methode | Volksgezondheidenzorg.info', Accessed 30 dec. https://www.volksgezondheidenzorg.info/onderwerp/kosten-van-ziekten/methode#node-verschillende-definities-van-zorguitgaven.

———. 2019c. 'Ziekte van Parkinson | Kosten | Zorguitgaven | Volksgezondheidenzorg.info', Accessed 30 dec. https://www.volksgezondheidenzorg.info/onderwerp/ziekte-van-parkinson/kosten/zorguitgaven#!node-zorguitgaven-ziekte-van-parkinson-naar-sector.

Nattinger, M. C., K. Mueller, F. Ullrich, and X. Zhu. 2016. 'Financial Performance of Rural Medicare ACOs', *J Rural Health,* Aug 24.

Neuwirth, E. B., J. Bellows, A. H. Jackson, and P. M. Price. 2012. 'How Kaiser Permanente uses video ethnography of patients for quality improvement, such as in shaping better care transitions', *Health Aff (Millwood),* 31: 1244-50.

Nicholas, L. H., J. B. Dimick, and T. J. Iwashyna. 2011. 'Do hospitals alter patient care effort allocations under pay-for-performance?', *Health Services Research,* 46: 61-81.

Nicholson, S., M. V. Pauly, A. Y. Wu, J. F. Murray, S. M. Teutsch, and M. L. Berger. 2008. 'Getting real performance out of pay-for-performance', *Milbank Q,* 86: 435-57.

Nicoletti, A., V. Sofia, A. Bartoloni, F. Bartalesi, H. Gamboa Barahon, S. Giuffrida, and A. Reggio. 2003. 'Prevalence of Parkinson's disease: a door-to-door survey in rural Bolivia', *Parkinsonism Relat Disord,* 10: 19-21.

Nijkrake, M. J., S. H. Keus, R. A. Oostendorp, S. Overeem, W. Mulleners, B. R. Bloem, and M. Munneke. 2009. 'Allied health care in Parkinson's disease: referral, consultation, and professional expertise', *Mov Disord,* 24: 282-6.

Nijkrake, M. J., S. H. Keus, S. Overeem, R. A. Oostendorp, T. P. Vlieland, W. Mulleners, E. M. Hoogerwaard, B. R. Bloem, and M. Munneke. 2010. 'The ParkinsonNet concept: development, implementation and initial experience', *Mov Disord,* 25: 823-9.

Norris, Pippa. 2001. *Digital divide : civic engagement, information poverty, and the Internet worldwide* (Cambridge University Press: Cambridge ; New York).

Olanow, C. W., K. Kieburtz, P. Odin, A. J. Espay, D. G. Standaert, H. H. Fernandez, A. Vanagunas, A. A. Othman, K. L. Widnell, W. Z. Robieson, Y. Pritchett, K. Chatamra, J. Benesh, R. A. Lenz, A. Antonini, and Lcig Horizon Study Group. 2014. 'Continuous intrajejunal infusion of levodopa-carbidopa intestinal gel for patients with advanced Parkinson's disease: a randomised, controlled, double-blind, double-dummy study', *Lancet Neurol,* 13: 141-9.

Oliver, D., S. Veronese, R.S. Morrison, H.I. Hurtig, and A.F. Eichler. 2019. 'Palliative approach to Parkinson disease and parkinsonian disorders', UpToDate, Accessed 06 march.

Orszag, P. R., and P. Ellis. 2007. 'The challenge of rising health care costs. A view from the Congressional Budget Office', *N Engl J Med,* 357: 1793-5.

Parkinson's Foundation. 2019a. 'Center of Excelence Network', Parkinson's Foundation, Accessed 09 may. https://parkinson.org/expert-care/centers-of-excellence.

———. 2019b. 'Expert Care Programs', Parkinson's Foundation, Accessed 09 may. https://parkinson.org/expert-care/Expert-Care-Programs.

Parkinson's UK. 2019. 'Improving the Quality of Parkinson's Healthcare', Accessed 15 sept. https://www.parkinsons.org.uk/about-us/improving-quality-parkinsons-healthcare.

Parkinson, J. 2002. 'An essay on the shaking palsy. 1817', *J Neuropsychiatry Clin Neurosci,* 14: 223-36; discussion 22.

ParkinsonNet. 2014. 'Radboudumc en Kaiser Permanente werken samen aan betere zorg voor parkinson', ParkinsonNet, Accessed 5 february. https://www.parkinsonnet.nl/nieuws/radboudumc-en-kaiser-permanente-werken-samen-aan-betere-zorg-voor-parkinson.

———. 2016. 'ParkinsonNet wordt ook in Noorwegen geïmplementeerd', PArkinosnNet, Accessed 05 februari. https://www.parkinsonnet.nl/nieuws/parkinsonnet-wordt-ook-in-noorwegen-geïmplementeerd.

———. 2019. 'optimale zorg voor Parkinson, dat is ons doel!', ParkinsonNet, Accessed 5 februari. https://www.parkinsonnet.nl/.

Pasluosta, C. F., H. Gassner, J. Winkler, J. Klucken, and B. M. Eskofier. 2015. 'An Emerging Era in the Management of Parkinson's Disease: Wearable Technologies and the Internet of Things', *IEEE J Biomed Health Inform,* 19: 1873-81.

Pearl, R. 2014. 'Kaiser Permanente Northern California: current experiences with internet, mobile, and video technologies', *Health Aff (Millwood),* 33: 251-7.

Perrin, A., and M. Duggan. June 26, 2015. 'Americans' Internet Access: 2000 - 2015', Pew Research Center, Accessed April 5. http://www.pewinternet.org/2015/06/26/americans-internet-access-2000-2015/.

Picillo, M., A. Nicoletti, V. Fetoni, B. Garavaglia, P. Barone, and M. T. Pellecchia. 2017. 'The relevance of gender in Parkinson's disease: a review', *J Neurol,* 264: 1583-607.

Pollack, C. E., K. W. Lemke, E. Roberts, and J. P. Weiner. 2015. 'Patient sharing and quality of care: measuring outcomes of care coordination using claims data', *Med Care,* 53: 317-23.

Pollack, CE., GE. Weissman, KW. Lemke, PS. Hussey, and JP. Weiner. 2012. 'Patient Sharing Among Physicians and Costs of Care: A Network Analytic Approach to Care Coordination Using Claims Data', *Journal of General Internal Medicine,* 28: 459-65.

Pollack, Craig Evan, Kevin D. Frick, Robert J. Herbert, Amanda L. Blackford, Bridget A. Neville, Antonio C. Wolff, Michael A. Carducci, Craig C. Earle, and Claire F. Snyder. 2014. 'It's who you know: patient-sharing, quality, and costs of cancer survivorship care', *Journal of cancer survivorship : research and practice,* 8: 156-66.

Porter, M. E., and E. O. Teisberg. 2006. *Redefining Health Care* (Harvard Business School Press: Boston, Massachusetts).

Power, A., and D. Ashby. 2014. 'Haemodialysis: hospital or home?', *Postgrad Med J,* 90: 92-7.

Qiang, J. K., and C. Marras. 2015. 'Telemedicine in Parkinson's disease: A patient perspective at a tertiary care centre', *Parkinsonism Relat Disord,* 21: 525-8.

Radder, D. L., N. M. de Vries, N. P. Riksen, S. J. Diamond, D. Gross, D. R. Gold, J. Heesakkers, E. Henderson, A. L. Hommel, H. H. Lennaerts, J. Busch, R. E. Dorsey, J. Andrejack, and B. R. Bloem. 2018. 'Multidisciplinary care for people with Parkinson's disease: the new kids on the block!', *Expert Rev Neurother.*

Radder, D. L. M., H. H. Lennaerts, H. Vermeulen, T. van Asseldonk, C. C. S. Delnooz, R. H. Hagen, M. Munneke, B. R. Bloem, and N. M. de Vries. 2020. 'The cost-effectiveness of specialized nursing interventions for people with Parkinson's disease: the NICE-PD study protocol for a randomized controlled clinical trial', *Trials,* 21: 88.

Ramirez, A. G., M. C. Tracci, G. J. Stukenborg, F. E. Turrentine, B. D. Kozower, and R. S. Jones. 2016. 'Physician-Owned Surgical Hospitals Outperform Other Hospitals in Medicare Value-Based Purchasing Program', *J Am Coll Surg,* 223: 559-67.

Rizzo, G., M. Copetti, S. Arcuti, D. Martino, A. Fontana, and G. Logroscino. 2016. 'Accuracy of clinical diagnosis of Parkinson disease: A systematic review and meta-analysis', *Neurology,* 86: 566-76.

Roland, M. 2012. 'Pay-for-performance: not a magic bullet.', *Ann Intern Med.,* 157: 912-3.

Roland, M., and S. Campbell. 2014. 'Successes and failures of pay for performance in the United Kingdom', *N Engl J Med,* 370: 1944-9.

Rosenthal, M. B., and R. A. Dudley. 2007. 'Pay-for-performance: will the latest payment trend improve care?', *JAMA,* 297: 740-4.

Roughead, E. E., S. J. Semple, and E. Rosenfeld. 2016. 'The extent of medication errors and adverse drug reactions throughout the patient journey in acute care in Australia', *Int J Evid Based Healthc,* 14: 113-22.

Ryan, A. M. 2009. 'Effects of the premier hospital quality incentive demonstration on medicare patient mortality and cost: Quality and performance', *Health Services Research,* 44: 821-42.

Ryan, A. M. , B. K. Nallamothu, and J. B. Dimick. 2012. 'Medicare's public reporting initiative on hospital quality had modest or no impact on mortality from three key conditions.', *Health Aff (Millwood),* 31: 585-92.

Ryan, A. M., J. Blustein, T. Doran, D. Michelow M, and L. P. Casalino. 2012. 'The effect of phase 2 of the premier hospital quality incentive demonstration on incentive payments to hospitals caring for disadvantaged patients', *Health Services Research,* 47: 1418-36.

Ryan, A. M., J. F. Burgess, M. F. Pesko, W. B. Borden, and J. B. Dimick. 2015. 'The early effects of Medicare's mandatory hospital pay-for-performance program', *Health Services Research,* 50: 81-97.

Ryan, A. M., S. Krinsky, E. Kontopantelis, and T. Doran. 2016. 'Long-term evidence for the effect of pay-for-performance in primary care on mortality in the UK: a population study', *Lancet,* 388: 268-74.

Safarpour, D., D. P. Thibault, C. L. DeSanto, C. M. Boyd, E. R. Dorsey, B. A. Racette, and A. W. Willis. 2015. 'Nursing home and end-of-life care in Parkinson disease', *Neurology,* 85: 413-9.

Santos-Garcia, D., and R. de la Fuente-Fernandez. 2015. 'Factors contributing to caregivers' stress and burden in Parkinson's disease', *Acta Neurologica Scandinavica,* 131: 203-10.

Saunders-Pullman, R., C. Wang, K. Stanley, and S. B. Bressman. 2011. 'Diagnosis and referral delay in women with Parkinson's disease', *Gend Med,* 8: 209-17.

Savica, R., B. R. Grossardt, J. H. Bower, J. E. Ahlskog, and W. A. Rocca. 2016. 'Time Trends in the Incidence of Parkinson Disease', *JAMA Neurol,* 73: 981-9.

Scott, B., A. Borgman, H. Engler, B. Johnels, and S.M. Aquilonius. 2000. 'Gender differences in Parkinson's disease symptom profile', *Acta Neurol Scand.,* 102: 37-43.

Serumaga, B., D. Ross-Degnan, A. J. Avery, R. A. Elliott, S. R. Majumdar, F. Zhang, and S. B. Soumerai. 2011. 'Effect of pay for performance on the management and outcomes of hypertension in the United Kingdom: Interrupted time series study', *BMJ,* 342: 322.

Shah, S. M., I. M. Carey, T. Harris, S. DeWilde, and D. G. Cook. 2011. 'Quality of chronic disease care for older people in care homes and the community in a primary care pay for performance system: Retrospective study', *BMJ,* 342: 587.

Shakir, M., K. Armstrong, and J.H. Wasfy. 2018. 'Could Pay-for-Performance Worsen Health Disparities?', *Health Policy,* 33: 567-69.

Sharp, A. L., Z. Song, D. G. Safran, M. E. Chernew, and A. Mark Fendrick. 2013. 'The effect of bundled payment on emergency department use: Alternative quality contract effects after year one', *Academic Emergency Medicine,* 20: 961-64.

Shih, T., and J. B. Dimick. 2013. 'Does pay-for-performance improve surgical outcomes? Evaluation of phase 2 of the premier hospital quality incentive demonstration project', *Journal of Surgical Research*, 179.

Shlebak, A., P. Sandhu, V. Ali, G. Jones, and C. Baker. 2016. 'The impact of the DoH Commissioning for Quality and Innovation incentive on the success of venous thromboembolism risk assessment in hospitalised patients. A single institution experience in a quality outcome improvement over a 4-year cycle', *JRSM Open,* 7: 2054270416632702.

Simpson, C. R., P. C. Hannaford, L. D. Ritchie, A. Sheikh, and D. Williams. 2011. 'Impact of the pay-for-performance contract and themanagement of hypertension in Scottish primary care: A 6-year population-based repeated cross-sectional study', *British Journal of General Practice,* 61: 443-51.

Skinner, T. R., I. A. Scott, and J. H. Martin. 2016. 'Diagnostic errors in older patients: a systematic review of incidence and potential causes in seven prevalent diseases', *Int J Gen Med,* 9: 137-46.

Smith, A. . 2014. 'Older Adults and Technology Use: Adoption is increasing, but many seniors remain isolated from digital life', Pew Research Center: Internet, Science & Tech, Accessed Jun 9. http://www.pewinternet.org/files/2014/04/PIP_Seniors-and-Tech-Use_040314.pdf.

Solla, P., A. Cannas, F.C. Ibba, F. Loi, M. Corona, G. Orofino, M.G. Marrosu, and F. Marrosu. 2012. 'Gender differences in motor and non-motor symptoms among Sardinian patients with Parkinson's disease', *J Neurol Sci.,* 323: 33-39.

Song, Z., D. G. Safran, B. E. Landon, Y. He, R. P. Ellis, R. E. Mechanic, M. P. Day, and M. E. Chernew. 2011. 'Health care spending and quality in year 1 of the alternative quality contract', *New England Journal of Medicine,* 365: 909-18.

Song, Z., D. G. Safran, B. E. Landon, M. B. Landrum, Y. He, R. E. Mechanic, M. P. Day, and M. E. Chernew. 2012. 'The 'alternative quality contract,' based on a global budget, lowered medical spending and improved quality', *Health Affairs,* 31: 1885-94.

Song, Z., D. G. Safran, B. E. Landon, S. Rose, M. Day, and M. E. Chernew. 2014. 'Payment reform in massachusetts: Effect of global payment on health care spending and quality 4 years into the alternative quality contract', *Journal of General Internal Medicine,* 29: S169.

Spehar, A. M., Campbell R. R., C. Cherrie, P. Palacios, D. Scott, J. L. Baker, B. Bjornstad, and J. Wolfson. 2005. 'Seamless Care: Safe Patient Transitions from Hospital to Home.' in K. Henriksen, J. B. Battles, E. S. Marks and D. I. Lewin (eds.), *Advances in Patient Safety: From Research to Implementation (Volume 1: Research Findings)* (Agency for Healthcare Research and Quality (US): Rockville (MD)).

Spindler, M.A., D. Tarsy, H.I. Hurtig, and A.F. Eichler. 2019. 'Initial pharmacologic treatment of Parkinson disease', UpToDate, Accessed 06 march.

Stocchi, F., and B R. Bloem. 2013. 'Move for Change Part II: a European survey evaluating the impact of the EPDA Charter for people with Parkinson's disease', *European Journal of Neurology,* 20: 461-72.

Strong, M., G. South, and R. Carlisle. 2009. 'The UK Quality and Outcomes Framework pay-for-performance scheme and spirometry: Rewarding quality or just quantity? A cross-sectional study in Rotherham, UK', *BMC Health Services Research,* 9.

Stuart, E. A., C. L. Barry, J. M. Donohue, S. F. Greenfield, K. Duckworth, Z. Song, R. Mechanic, E. M. Kouri, C. Ebnesajjad, M. E. Chernew, and H. A. Huskamp. 2016. 'Effects of accountable care and payment reform on substance use disorder treatment: evidence from the initial 3 years of the alternative quality contract', *Addiction,* Aug 12.

Sturkenboom, I. H., M. J. Graff, J. C. Hendriks, Y. Veenhuizen, M. Munneke, B. R. Bloem, and M. W. Nijhuis-van der Sanden. 2014. 'Efficacy of occupational therapy for patients with Parkinson's disease: a randomised controlled trial', *Lancet Neurol,* 13: 557-66.

Sturkenboom, I. H., J. C. Hendriks, M. J. Graff, E. M. Adang, M. Munneke, M. W. Nijhuis-van der Sanden, and B. R. Bloem. 2015. 'Economic evaluation of occupational therapy in Parkinson's disease: A randomized controlled trial', *Mov Disord,* 30: 1059-67.

Sturkenboom, I.H.W.M., M.C.E. Thijssen, J.J. Gons-van Elsacker, I.J.H. Jansen, A. Maasdam, M. Schulten, D. Vijver-Visser, E.J.M. Steultjens, B.R. Bloem, and M. Munneke. 2008. "Guidelines for Occupational Therapy in Parkinson's Disease Rehabilitation." In. Nijmegen, The Netherlands/Miami (FL), U.S.A.:: ParkinsonNet/NPF.

Tai, W., L. Kalanithi, and A. Milstein. 2014. 'What can be achieved by redesigning stroke care for a value-based world?', *Expert Rev Pharmacoecon Outcomes Res,* 14: 585-7.

Tan, Q., Z. J. Hildon, S. Singh, J. Jing, T. L. Thein, R. Coker, H. J. M. Vrijhoef, and Y. S. Leo. 2017. 'Comparing patient and healthcare worker experiences during a dengue outbreak in Singapore: understanding the patient journey and the introduction of a point-of-care test (POCT) toward better care delivery', *BMC Infect Dis,* 17: 503.

Thrift-Perry, M., A. Cabanes, F. Cardoso, K. M. Hunt, T. A. Cruz, and K. Faircloth. 2018. 'Global analysis of metastatic breast cancer policy gaps and advocacy efforts across the patient journey', *Breast*, 41: 93-106.

Tian, M., V. S. Ajay, D. Dunzhu, S. S. Hameed, X. Li, Z. Liu, C. Li, H. Chen, K. Cho, R. Li, X. Zhao, D. Jindal, I. Rawal, M. K. Ali, E. D. Peterson, J. Ji, R. Amarchand, A. Krishnan, N. Tandon, L. Q. Xu, Y. Wu, D. Prabhakaran, and L. L. Yan. 2015. 'A Cluster-Randomized, Controlled Trial of a Simplified Multifaceted Management Program for Individuals at High Cardiovascular Risk (SimCard Trial) in Rural Tibet, China, and Haryana, India', *Circulation,* 132: 815-24.

Tompkins C, Higgins A, Perloff J, Veselovskiy G, . 2013. "Population Health Management In Medicare Advantage." In. Health Affairs Blog.

Tysnes, O. B., and A. Storstein. 2017. 'Epidemiology of Parkinson's disease', *J Neural Transm (Vienna)*, 124: 901-05.

Vaghela, P., M. Ashworth, P. Schofield, and M. C. Gulliford. 2009. 'Population intermediate outcomes of diabetes under pay-for-performance incentives in England from 2004 to 2008', *Diabetes Care,* 32: 427-29.

van Asseldonk, M.J.M.D., H.C. Dicke, B.J.W. van den Beemt, D.J. van den Berg, S. ter Borg, G.M. Duin, S.L.H.M. Govers, J.J. van Teeffelen, J.I. Hoff, B. van Harten, B.R. Bloem, S.H.J. Keus, and M. Munneke. 2012. "Dietetics guideline for Parkinson's disease." In. The Hague, The Netherlands.

Van Den Eeden, S. K., C. M. Tanner, A. L. Bernstein, R. D. Fross, A. Leimpeter, D. A. Bloch, and L. M. Nelson. 2003. 'Incidence of Parkinson's disease: variation by age, gender, and race/ethnicity', *Am J Epidemiol,* 157: 1015-22.

van der Eijk, M., B. R. Bloem, F. A. Nijhuis, J. Koetsenruijter, H. J. Vrijhoef, M. Munneke, M. Wensing, and M. J. Faber. 2015. 'Multidisciplinary Collaboration in Professional Networks for PD A Mixed-Method Analysis', *J Parkinsons Dis,* 5: 937-45.

van der Eijk, M., M. J. Faber, S. Al Shamma, M. Munneke, and B. R. Bloem. 2011. 'Moving towards patient-centered healthcare for patients with Parkinson's disease', *Parkinsonism Relat Disord,* 17: 360-4.

van der Eijk, M., M. J. Faber, B. Post, M. S. Okun, P. Schmidt, M. Munneke, and B. R. Bloem. 2015. 'Capturing patients' experiences to change Parkinson's disease care delivery: a multicenter study', *J Neurol,* 262: 2528-38.

van der Maaten, L., and G. Hinton. 2008. 'Visualizing Data using t-SNE', *Journal of Machine Learning Research,* 9: 2579-605.

van der Marck, M. A., B. R. Bloem, G. F. Borm, S. Overeem, M. Munneke, and M. Guttman. 2013. 'Effectiveness of multidisciplinary care for Parkinson's disease: a randomized, controlled trial', *Mov Disord,* 28: 605-11.

van der Marck, M. A., M. Munneke, W. Mulleners, E. M. Hoogerwaard, G. F. Borm, S. Overeem, and B. R. Bloem. 2013. 'Integrated multidisciplinary care in Parkinson's disease: a non-randomised, controlled trial (IMPACT)', *Lancet Neurol,* 12: 947-56.

van Halteren, A. D., M. Munneke, E. Smit, S. Thomas, B. R. Bloem, and S. K. L. Darweesh. 2020. 'Personalized Care Management for Persons with Parkinson's Disease', *J Parkinsons Dis,* 10: S11-s20.

van Herck, P., D. de Smedt, L. Annemans, R. Remmen, M. B. Rosenthal, and W. Sermeus. 2010. 'Systematic review: effects, design choices, and context of pay-for-performance in health care', *BMC Health Services Research,* 10: 247.

Vektis. 2017. "overleg 29-9-2017." In, edited by F.P. Vlaanderen. Vektis.

———. 2018. 'Over Vektis', Vektis, Accessed 10-1-2019. https://www.vektis.nl/over-vektis.

———. 2020. 'Over Vektis', Vektis, Accessed 10-08-2020. https://www.vektis.nl/over-vektis.

Vlaanderen, F. P., Y. de Man, J. H. Krijthe, M. A. C. Tanke, A. S. Groenewoud, P. P. T. Jeurissen, S. Oertelt-Prigione, M. Munneke, B. R. Bloem, and M. J. Meinders. 2019. 'Sex-Specific Patient Journeys in Early Parkinson's Disease in the Netherlands', *Front Neurol,* 10: 794.

Vlaanderen, F.P., P.P.T. Jeurissen, M.J. Faber, M. Munneke, and B.R. Bloem. 2016. 'The Dutch outcome-based payment model of ParkinsonNet: a case study', *Paris*: In print.

Wang, Y., C. A. O'Donnell, D. F. Mackay, and G. C. M. Watt. 2006. 'Practice size and quality attainment under the new GMS contract: A cross-sectional analysis', *British Journal of General Practice,* 56: 830-35.

Wensing, M., M. van der Eijk, J. Koetsenruijter, B. R. Bloem, M. Munneke, and M. Faber. 2011. 'Connectedness of healthcare professionals involved in the treatment of patients with Parkinson's disease: a social networks study', *Implement Sci,* 6: 67.

Werner, R. M., and R. A. Dudley. 2009. 'Making the 'pay' matter in pay-for-performance: implications for payment strategies.', *Health Aff (Millwood).* 28: 1498-508.

Werner, R. M., J. T. Kolstad, E. A. Stuart, and D. Polsky. 2011. 'The effect of pay-for-performance in hospitals: lessons for quality improvement', *Health Aff.*, 30: 690-8.

Whalley, D., H. Gravelle, and B. Sibbald. 2008. 'Effect of the new contract on GPs' working lives and perceptions of quality of care: A longitudinal survey', *British Journal of General Practice,* 58: 8-14.

Willis, A. W., M. Schootman, B. A. Evanoff, J. S. Perlmutter, and B. A. Racette. 2011. 'Neurologist care in Parkinson disease: a utilization, outcomes, and survival study', *Neurology*, 77: 851-7.

Willis, A. W., M. Schootman, R. Tran, N. Kung, B. A. Evanoff, J. S. Perlmutter, and B. A. Racette. 2012. 'Neurologist-associated reduction in PD-related hospitalizations and health care expenditures', *Neurology,* 79: 1774-80.

Willis, Cameron D., Barbara L. Riley, Carol P. Herbert, and Allan Best. 2013. 'Networks to strengthen health systems for chronic disease prevention', *American journal of public health,* 103: e39-e48.

Wilson, L. S., and A. J. Maeder. 2015. 'Recent Directions in Telemedicine: Review of Trends in Research and Practice', *Healthc Inform Res,* 21: 213-22.

Worlds Parkinson's Program. 2019. 'About', Accessed 15 sept. https://www.pdprogram.org/about-us/.

Xu, K., A. Soucat, Kutzin J., C. Brindley, N. Vande Maele, H. Touré, M. Aranguren-Garcia, D. Li, H. Barroy, B. Flores, T. Roubal, C. Indikadahena, V. Cherilova, and A. Siroka. 2018. "Public Spending on Health: A Closer Look at Global Trends." In. Geneva: WHO.

Yang, J. H., S. M. Kim, S. J. Han, M. Knaak, G. H. Yang, K. D. Lee, Y. H. Yoo, G. Ha, E. J. Kim, and M. S. Yoo. 2016. 'The impact of Value Incentive Program (VIP) on the quality of hospital care for acute stroke in Korea', *International Journal for Quality in Health Care,* 28: 580-85.

Ypinga, J. H. L., N. M. de Vries, Lhhm Boonen, X. Koolman, M. Munneke, A. H. Zwinderman, and B. R. Bloem. 2018a. 'Effectiveness and costs of specialised physiotherapy given via ParkinsonNet: a retrospective analysis of medical claims data', *Lancet Neurol,* 17: 153-61.

Zhao, M., D. R. Haley, A. Spaulding, and H. A. Balogh. 2015. 'Value-based purchasing, efficiency, and hospital performance', *Health Care Manag.*, 34: 4-13.

Zorgwijzer. 2019. "Cijfers en feiten over de zorgverzekering." In. Barendrecht, the Netherlands: Zorgwijzer.

Summary

Chapter 1

Parkinson's disease (PD), is a chronic progressive neurodegenerative disorder, characterized by a wide range of symptoms. Many different healthcare providers are involved in the treatment of people with PD, at substantial costs. To achieve optimal outcomes, care for people with PD needs to be seamless and sustainable. In this thesis we used the following definitions: *seamless care* is care which is consistent and coherent, marked by an orderly, logically and aesthetically consistent relation of parts, without discontinuities or disparities, uniform in quality and combined in an inconspicuous way. *Sustainable care* is care which can be sustained through time, and that is efficient and does not unnecessary drain finite resources.

Despite efforts, the current care for people with PD is often far from seamless and sustainable. First, the care people with PD receive is quite variable, which implies that care is not 'consistent', 'coherent' or 'uniform in quality'. Second, people with PD do not regard their care as 'marked by an orderly, logically and aesthetically consistent relation of parts'. Third, financial issues in healthcare systems stand in the way of seamless and sustainable care delivery. And finally, care for people with PD is delivered by scattered healthcare providers, complicating multidisciplinary collaboration, and as a result, healthcare providers treat few people with PD, and do not become experts.

The aim of this thesis is to study potential measures to achieve seamless and sustainable care for people with PD. This results in the following research questions: How does the current patient journey of people with PD look like? What are the current seams in the care for people with PD? Can outcome-based payment models contribute to seamless and sustainable care for people with PD? Will care for people with PD be more seamless and sustainable if providers share more patients?

Chapter 2

In Chapter 2 we used the Dutch national medical claims database to reconstructed the sex-specific patient journey for PD during the first five years after diagnosis. In a time-to-event analysis using claims data of all persons diagnosed with PD between 2012 and 2016 in the Netherlands, we identified the moments in time when these persons received care from neurologists, allied healthcare therapists and general practitioners. Similarly, we identified the time points at which relevant clinical milestones were reached: the occurrence of PD-related complications (e.g. pneumonia, orthopaedic injury, and PD-related hospitalization), nursing home admission, and mortality.

The analysis included claims data of 13,518 men and 8,775 women with newly diagnosed PD. It appeared that after diagnosis, women visited on average their general practitioner 10 days and the physiotherapist 89 days sooner than men. We found little difference in this regard for neurologist utilization. Approximately two years after diagnosis, the first PD-related complication occurred (women: 1.8 years; men: 2.3 years). Within five years, 37.9% of the women had at least once visited an occupational therapist and 18.5% a speech & language therapist. The corresponding figures for men were 33.1% and 23.7%. Furthermore, 72.9% of women and 68.7% of men had experienced at least one PD-related complication; 27.5% of women and 22.5% of men had been admitted to a nursing home. Within five years after diagnosis, 14.6% of women and 18.3% of men had died. These sex differences might be due to a doctor or patient delay in the diagnostic process in women, rather than to a more aggressive disease progression in women.

Chapter 3

In Chapter 3 we identified the core needs of people with PD using the Voice of the Customer (VoC) approach: this is a novel methodology originally developed in the field of industry to probe the clients' needs. A group of 12 interviewers carried out in-depth interviews with people with PD (n=20), relatives (n=12) and healthcare providers (n=11). The interviewers identified ten frequently mentioned needs and combined the most informative quotations into a comprehensive video, which was shown to the interviewees in a consensus meeting. During that meeting, the interviewees prioritized the ten identified needs by allocating a maximum of three points to each of the different needs. They could allocate all three points to one need or divide these between two or three needs. The total number of points represented the importance of the topic.

The VoC approach revealed that the most urgent patient needs concerned the social, emotional or domestic domain, rather than the bio-medical aspects of the disease. The top unmet needs were: (1) more self management; (2) better interdisciplinary collaboration between different healthcare providers; (3) more time to discuss the future and possible scenarios; and (4) a healthcare provider acting as personal case manager, either to solve problems directly or to direct people with PD to the professional best equipped to address the problem at hand. These results may serve to optimize the care for people with PD.

Chapter 4

Chapter 4 describes a review on the effects of outcome-based payment models (OBPMs) on quality of care and healthcare costs. We first developed a definition for OBPMs: a payment model in healthcare in which the performance-related incentive payments for the healthcare providers depend for at least 10% on outcomes of the provided care, and which is designed to stimulate favourable effects in terms of quality of care or healthcare costs. Next, we searched four data sources to identify the models:

1) scientific literature databases; 2) websites of relevant governmental and scientific agencies; 3) the reference lists of included articles; 4) experts in the field. We selected only studies that examined the impact of the payment model on quality and/or costs. A narrative evidence synthesis was used to link specific design features to effects on quality of care or healthcare costs.

We included 88 articles, describing 12 OBPMs. None of them specifically focused on PD; though all models targeted populations with chronic conditions. Based on differences in design features, we could distinguish two groups of OBPMs: narrow OBPMs, which contain only explicit financial incentives for objectively measured quality performance; and broad OBPMs, which combine global budgets and risk sharing for multidisciplinary provider groups with explicit financial incentives for quality. Most (5 out of 9) narrow OBPMs showed positive effects on quality; the others had mixed (2) or negative (2) effects. The effects of narrow OBPMs on healthcare utilization or costs, however, were unfavourable (3) or unknown (6). The three broad OBPMs showed all positive effects on quality of care, while reducing healthcare cost growth. Although only three broad OBPMs could be included in this review, these were the ones that had the most favourable effects on both quality of care and healthcare utilization/costs.

Chapter 5

In chapter 5, we noted that people with PD create so called 'patient-sharing networks' of healthcare providers by choosing their own healthcare providers. These networks differ in 'density': the number of patients the providers share. Our objective was to identify whether denser healthcare provider networks could be linked to better patient outcomes, lower healthcare utilization and lower healthcare costs in PD. We used claims data of all people with PD in the Netherlands between 2012 and 2016. We included all claims of those healthcare providers most frequently involved in care for people with PD. To assess the density of healthcare provider networks, we used the average number of patients that healthcare providers share as a proxy for the density of their collaboration (density score). We calculated the average density score per person with PD and per hospital population. We visualized collaboration graphs of the top and bottom three hospitals using t-distributed Stochastic Neighbour Embedding (t-SNE). Using logistic and linear regression analyses, we estimated the relationship between density and health outcomes, healthcare utilization, and healthcare costs.

The average density score varied considerably on the patient level (average 6.7, SD 8.2), as well as on the level of hospital populations (average 3.9, SD 8.2). Adjusted for confounders, higher density scores were associated with a lower risk of PD-related complications and with lower healthcare costs, but not with mortality. Higher density scores were associated with more frequent involvement of neurologists, physiotherapists and occupational therapists, but less frequent involvement of psychologists. We therefore propose to use density scores as a quality indicator for provider networks.

Chapter 6

In Chapter 6, we discussed how we could improve the care for people with PD by moving it to the patient's home. Care of individuals with PD is often poorly designed: individuals with PD who are hospitalized often experience delayed treatment, contra-indicated medications, prolonged immobility, lengthy stays, and high mortality. Despite the documented benefits of receiving care from clinicians with PD expertise, many people with PD do not receive such expert care. Moreover, current care models frequently require older individuals with impaired mobility, cognition, and driving ability to be driven by overburdened informal caregivers to large, complex urban medical centers.

Moving care to the patient's home would make PD care more patient-centered. Demographic factors, including aging of the populations, and social factors such as the splintering of the extended family, will increase the need for home-based care. Technological advances, especially the ability to assess and deliver care remotely, will enable the transition of care back to the home situation. However, despite its promise, this next generation of home-based care will have to overcome barriers, including outdated insurance models and a technological divide. Once addressed, home-based care will increase access to high-quality care for the growing number of individuals with PD.

Chapter 7

In Chapter 7 we interpreted our findings and formed our conclusions. We started with an overview of the main findings. First, the reconstruction of the Parkinson patient's journey during the first five years after diagnosis revealed profound sex differences: women visit most of the included healthcare providers sooner after diagnosis and more frequently than do men (Chapter 2). Second, patients' top unmet needs were: 1) more self management; 2) better interdisciplinary collaboration between different healthcare providers; 3) more time to discuss the future and possible scenarios; and 4) a healthcare provider who can act as a personal case manager (Chapter 3). Third, broad OBPMs seem to have the most promising effects on quality of care and healthcare costs (Chapter 4). Fourth, denser networks are associated with lower occurrence of PD-related complications, higher utilization, but lower costs (Chapter 5). Finally, care needs to be offered closer to the patient's home (Chapter 6).

Our interpretations are the following. Care for people with PD in the Netherlands is currently neither seamless nor sustainable. The seams we identified all concern organisational aspects, rather than bio-medical aspects. The identified seam of

unexplained sex-differences in healthcare use might be due to doctor's or patient delay in the diagnostic process in women. The most urgent seams reported by people with PD themselves concern the social, emotional or domestic domain. To identify seams in care, the perspective of the individual people with PD needs to be included, and the Voice of the Customer method might be an aid to this.

We offered some suggestions on how to address these seams. First, the way in which providers are organised and have divided tasks needs to be rethought. We argue that specialist Parkinson's nurses are best suited to act as a personal case manager. Second, video visits and self-monitoring apps can help to overcome some seams in PD care. Third, we would welcome the development of home-based care programs for PD, rather than the more hospital-based models of current care.

Concerning the payment model for PD care, we suggest an experimental broad OBPM that links financial incentives to quality indicators relevant for PD. 'Density' could therefore act as a quality indicator in this new model, whereby high density has favourable effects on quality of care and healthcare costs.

Summarizing the strengths and limitations of the research, we propose that applying a mix of research methodologies is a major strength. Other strengths are the large samples of our analyses and the comprehensiveness of our systematic review. One limitation is the lack of long-term effects; i.e., beyond five years after diagnosis. A second limitation is that our review on OBPMs is not PD-specific. And finally, the activities of ParkinsonNet might have influenced the generalisability of our findings in several chapters.

In our future outlook we advocate for more research on sex-specific doctor' or patient' delays in PD to explain the 'earlier' use of multiple health services in women with PD. We would recommend a pilot in which specialist PD nurses act as a personal case manager for people with PD, to confirm whether this can make care for people with PD more seamless. We would like to enhance the use of technological innovations such as self-monitoring apps. We suggest innovative care models that deliver care for people with PD closer at home. And finally, we advocate for the development of an OBPM for PD care with density as incorporated quality indicator. We encourage a pilot to study the effects of this model on quality of care and healthcare costs. A better, more seamless and more sustainable care delivery for PD can serve as a blueprint example for other chronic conditions.

Samenvatting

Hoofdstuk 1

De ziekte van Parkinson (ZvP), is een chronische progressieve neurodegeneratieve aandoening, gekenmerkt door een breed scala aan symptomen. Bij de behandeling van mensen met de ZvP zijn veel verschillende zorgaanbieders betrokken, en daar gaan aanzienlijke zorgkosten mee gepaard. Om optimale resultaten te bereiken, moet de zorg voor mensen met ZVP naadloos en betaalbaar zijn. In dit proefschrift hebben we de volgende definities gehanteerd: *naadloze zorg* is zorg die consistent en coherent is, gekenmerkt door een ordelijke, logisch en esthetisch consistente relatie van onderdelen, zonder discontinuïteiten of ongelijkheden, uniform in kwaliteit en gecombineerd op een natuurlijke manier. *Betaalbare zorg* is zorg waarbij de kwaliteit die door de tijd heen gehandhaafd kan blijven; die efficiënt is, en die niet onnodig (financiële) bronnen of mankracht uitput.

Ondanks inspanningen is de huidige zorg voor mensen met de ZvP vaak verre van naadloos en betaalbaar. Ten eerste is de zorg die mensen met de ZvP ontvangen nogal variabel, wat impliceert dat de zorg niet 'consistent', 'coherent' of 'uniform van kwaliteit' is. Ten tweede beschouwen mensen met de ZvP hun zorg niet als 'gekenmerkt door een ordelijke, logisch en esthetisch consistente relatie van onderdelen'. Ten derde staan financiële problemen in de gezondheidszorg een naadloze en betaalbare zorgverlening in de weg. En tot slot wordt de zorg voor mensen met de ZvP geleverd door zeer veel individuele zorgaanbieders. Dit bemoeilijkt de multidisciplinaire samenwerking, en belemmert expertise vorming omdat een individuele zorgaanbieder weinig mensen met de ZvP behandelt.

Het doel van dit proefschrift is om mogelijke maatregelen te bestuderen die naadloze en betaalbare zorg voor mensen met de ZvP dichterbij brengt. Dit levert de volgende onderzoeksvragen op: Hoe ziet de huidige patiëntenreis van mensen met de ZvP eruit? Wat zijn de huidige naden in de zorg voor mensen met de ZvP? Kunnen uitkomstbekostiningsmodellen (UBM's) bijdragen aan naadloze en betaalbare zorg voor mensen met de ZvP? Zal de zorg voor mensen met de ZvP naadlozer en beter betaalbaar zijn als zorgverleners meer gemeenschappelijke patiënten hebben?

Hoofdstuk 2

In Hoofdstuk 2 hebben we gebruik gemaakt van een landelijke database van zorgverzekeraars om de sekse-specifieke patiëntreis voor de ZvP gedurende de eerste vijf jaar na de diagnose te reconstrueren. Met een time-to-event analyse op de declaratiegegevens van alle mensen in Nederland bij wie de ZvP tussen 2012 en 2016 is vastgesteld, hebben we de momenten in kaart gebracht waarop deze personen zorg kregen van neurologen, paramedici en huisartsen. Ook hebben we de tijdstippen geïdentificeerd waarop relevante klinische mijlpalen werden bereikt: het optreden van ZvP-gerelateerde complicaties (bijv. Longontsteking, orthopedisch letsel en ZvP-gerelateerde ziekenhuisopname), verpleeghuisopnames en mortaliteit.

De analyse omvatte declaratiegegevens van 13.518 mannen en 8.775 vrouwen met nieuw gediagnosticeerde ZvP. Het bleek dat vrouwen na de diagnose hun huisarts gemiddeld 10 dagen eerder bezochten dan mannen en hun fysiotherapeut gemiddeld 89 dagen eerder. We vonden in dit opzicht weinig verschil voor het bezoek aan neurologen. Ongeveer twee jaar na de diagnose trad de eerste ZVP-gerelateerde complicatie op (bij vrouwen: na 1,8 jaar; bij mannen: na 2,3 jaar). Binnen vijf jaar had 37,9% van de vrouwen minstens één keer een ergotherapeut en 18,5% minstens één keer een logopedist bezocht. De overeenkomstige cijfers voor mannen waren 33,1% en 23,7%. Bovendien had 72,9% van de vrouwen en 68,7% van de mannen ten minste één ZvP-gerelateerde complicatie ervaren. 27,5% van de vrouwen en 22,5% van de mannen waren na vijf jaar opgenomen in een verpleeghuis. Binnen vijf jaar na de diagnose waren 14,6% van de vrouwen en 18,3% van de mannen overleden. Deze sekseverschillen lijken eerder te wijten zijn aan een doctor of patient delay in het diagnostische proces bij vrouwen, dan aan een agressievere ziekteprogressie bij vrouwen.

Hoofdstuk 3

In Hoofdstuk 3 hebben we de kernbehoeften van mensen met de ZvP geïdentificeerd met behulp van de Voice of the Customer (VoC) benadering: een nieuwe methodologie die oorspronkelijk in de industrie is ontwikkeld om de behoeften van klanten te onderzoeken. Een groep van 12 interviewers voerde diepte-interviews uit met mensen met de ZvP (n = 20), familieleden (n = 12) en zorgverleners (n = 11). De interviewers identificeerden 10 vaak genoemde behoeften en combineerden de meest informatieve citaten tot een uitgebreide videoboodschap, die tijdens een consensusvergadering aan de geïnterviewden werd getoond. Tijdens die bijeenkomst hebben de geïnterviewden prioriteit toegekend aan de tien geïdentificeerde behoeften door elk drie punten toe te wijzen aan een van de verschillende behoeften. Ze zouden alle drie de punten aan één behoefte kunnen toewijzen of deze over twee of drie behoeften kunnen verdelen. Het totale aantal punten representeerde zodoende het belang van het onderwerp.

Uit de VoC-benadering kwam naar voren dat de meest urgente behoeften van de patiënt betrekking hadden op het sociale, emotionele of huiselijke domein, en niet op de biomedische aspecten van de ziekte. De belangrijkste onvervulde behoeften waren: (1) meer eigen regie; (2) betere samenwerking tussen verschillende zorgaanbieders; (3) meer tijd om mogelijke toekomstscenario's te bespreken; en (4) een zorgverlener die optreedt als persoonlijke aanspreekpunt, hetzij om problemen rechtstreeks op te lossen, hetzij om mensen met de ZvP door te verwijzen naar de professional die het

best is toegerust om het probleem aan te pakken. Deze resultaten kunnen dienen om de zorg voor mensen met de ZvP te optimaliseren.

Hoofdstuk 4

Hoofdstuk 4 beschrijft een overzicht van de effecten van uitkomstbekostigingsmodellen (UBM's) op de kwaliteit van zorg en de zorgkosten. We ontwikkelden eerst een definitie voor UBM's: een bekostigingsmodel in de zorg waarbij kwaliteitsprikkels voor zorgaanbieders voor minimaal 10% afhankelijk zijn van de uitkomsten van de geleverde zorg, en dat gericht is op het stimuleren van gunstige kwaliteitseffecten van zorg of zorgkosten. Vervolgens hebben we vier databronnen doorzocht om deze modellen te identificeren: 1) wetenschappelijke literatuurdatabases; 2) websites van relevante overheids- en wetenschappelijke instanties; 3) de referentielijsten van opgenomen artikelen; 4) consultatie van experts. We selecteerden onderzoeken die de impact van het betalingsmodel op kwaliteit en/of kosten onderzochten. Een narratieve evidence-synthese werd gebruikt om specifieke ontwerpkenmerken van de modellen te koppelen aan de gevonden effecten op de kwaliteit van zorg en de zorgkosten.

We hebben 88 artikelen geïncludeerd die 12 UBM's beschrijven. Geen van de modellen was specifiek gericht op de ZvP; hoewel alle modellen gericht waren op chronische aandoeningen. Op basis van verschillen in ontwerpkenmerken konden we twee groepen UBM's onderscheiden: smalle UBM's, die alleen expliciete financiële prikkels bevatten voor objectief gemeten kwaliteitsindictoren; en brede UBM's, die globale budgetten en risicodeling voor multidisciplinaire zorgaanbieders combineren met expliciete financiële prikkels voor kwaliteit. De meeste (5 van de 9) smalle UBM's vertoonden positieve effecten op de kwaliteit; de anderen hadden gemengde (2) of negatieve (2) effecten. De effecten van smalle UBM's op het zorggebruik of de zorgkosten waren echter ongunstig (3) of onbekend (6). De drie brede UBM's lieten allen positieve effecten zien op de kwaliteit van zorg, terwijl de groei van de zorgkosten afnam. Hoewel in dit review slechts drie brede UBM's konden worden geïncludeerd, hadden deze de meest gunstige effecten op zowel kwaliteit van zorg als op het zorggebruik en de zorgkosten.

Hoofdstuk 5

In hoofdstuk 5 bestudeerden we de zogenoemde 'patient-sharing networks' van zorgaanbieders die mensen met de ZvP creëren door hun zorggebruik. Deze netwerken verschillen in 'dichtheid': het aantal gemeenschappelijke patiënten dat de zorgverleners hebben. Ons doel was om vast te stellen of dichtere netwerken kunnen worden gekoppeld aan betere zorguitkomsten, lager zorggebruik en lagere zorgkosten voor de ZVP. We hebben gebruik gemaakt van declaratiegegevens van alle mensen met de ZvP in Nederland tussen 2012 en 2016. We hebben alle declaraties meegenomen van de zorgaanbieders die het meest betrokken zijn bij zorg voor mensen met de ZvP.

Om de dichtheid van netwerken van zorgaanbieders te beoordelen, hebben we het gemiddelde aantal patiënten dat zorgaanbieders delen, gebruikt als een maatstaf voor de dichtheid van hun samenwerking (dichtheidsscore). We berekenden de gemiddelde dichtheidsscore per patiënt en per ziekenhuispopulatie. We visualiseerden deze dichtheidsscores van de hoogst scorende en de laagst scorende drie ziekenhuizen met behulp van t-gedistribueerde Stochastic Neighbor Embedding (t-SNE). Met behulp van logistieke en lineaire regressieanalyses hebben we de relatie geschat tussen dichtheidsscores enerzijds en de gezondheidsresultaten, zorggebruik en zorgkosten anderzijds.

De gemiddelde dichtheidsscore varieerde aanzienlijk op patiëntniveau (gemiddeld 6,7; SD 8,2), maar ook op het niveau van ziekenhuispopulaties (gemiddeld 3,9; SD 8,2). Gecorrigeerd voor confounders waren hogere dichtheidsscores geassocieerd met een lager risico op ZvP-gerelateerde complicaties en met lagere zorgkosten, maar niet met een lagere mortaliteit. Hogere dichtheidsscores werden geassocieerd met frequente betrokkenheid van neurologen, fysiotherapeuten en ergotherapeuten, maar met minder frequente betrokkenheid van psychologen. We stellen daarom voor om dichtheidsscores te gebruiken als kwaliteitsindicator voor zorgnetwerken.

Hoofdstuk 6

In Hoofdstuk 6 bespraken we hoe we de zorg voor mensen met de ZvP kunnen verbeteren door de zorg zo veel mogelijk bij de patiënt thuis te leveren. De zorg voor mensen met de ZvP is vaak slecht opgezet: mensen met de ZvP die in het ziekenhuis worden opgenomen, komen vaak in aanraking met ertraagde behandelingen, gecontra-indiceerde medicatie, langdurige immobiliteit, langdurige opnames en hoge mortaliteit. Ondanks de voordelen van het ontvangen van zorg door clinici met ZvP-expertise, ontvangen veel mensen met de ZvP dergelijke deskundige zorg niet. Bovendien vereisen de huidige zorgsystemen vaak dat oudere personen met beperkte mobiliteit, cognitie of rijvaardigheid door overbelaste mantelzorgers naar grote, complexe stedelijke medische centra worden gebracht.

Door de zorg naar het huis van de patiënt te verplaatsen, zou de Parkinsonzorg patiëntgerichter kunnen worden. Demografische factoren, waaronder vergrijzing van de bevolking, en sociale factoren zoals de verwatering van familiebanden, zullen de behoefte aan thuiszorg doen toenemen. Technologische vooruitgang, met name het vermogen om zorg op afstand te verlenen, zal de overgang van zorg naar de thuissituatie mogelijk maken. Ondanks bovenstaande zullen er barrières overwonnen moeten worden, waaronder verouderde verzekeringsmodellen en beperkte digitale vaardigheden van sommige patiënten. Eenmaal aangepakt, zal zorg thuis de toegang tot kwalitatief hoogwaardige zorg voor het groeiende aantal mensen met de ZvP vergroten.

Hoofdstuk 7

In Hoofdstuk 7 presenteren we de interpretatie van onze bevindingen en de conclusies die wij daaruit trekken. We zijn begonnen met een overzicht van de belangrijkste bevindingen. Ten eerste bracht de reconstructie van de reis van mensen met de ZvP gedurende de eerste vijf jaar na de diagnose grote sekseverschillen aan het licht: vrouwen bezoeken de meeste zorgverleners eerder en vaker dan mannen (Hoofdstuk 2). Ten tweede waren de belangrijkste onvervulde behoeften van mensen met de ZvP: 1) meer eigen regie; 2) betere samenwerking tussen verschillende zorgaanbieders; 3) meer gelegenheid om toekomstscenario's te bespreken; en 4) een zorgaanbieder die kan optreden als persoonlijk aanspreekpunt (Hoofdstuk 3). Ten derde lijken brede UBM's de meest belovende effecten te hebben op de kwaliteit van zorg en de zorgkosten (Hoofdstuk 4). Ten vierde worden dichtere netwerken geassocieerd met minder ZvP-gerelateerde complicaties, hogere zorggebruik, maar lagere zorgkosten (Hoofdstuk 5). Ten slotte moet de zorg dichter bij huis worden geleverd (Hoofdstuk 6).

Onze interpretaties zijn de volgende. De zorg voor mensen met de ZvP in Nederland is momenteel noch naadloos, noch betaalbaar. De naden die we identificeerden, hebben allemaal betrekking op organisatorische aspecten en niet op biomedische aspecten. De geïdentificeerde naad van onverklaarde sekseverschillen bij het gebruik van de gezondheidszorg kan te wijten zijn aan een doctor of patient delay in het diagnostische proces bij vrouwen. De meest urgente naden die mensen met ZVP zelf melden, betreffen het sociale, emotionele of huiselijke domein. Om naden in de zorg te identificeren, moet het perspectief van de individuele mensen met de ZvP worden meegenomen, en de Voice of the Customer-methode kan daarbij een hulpmiddel zijn.

We hebben enkele suggesties gedaan om deze naden aan te pakken. Ten eerste moet de manier waarop zorgaanbieders zijn georganiseerd opnieuw worden bekeken. Wij stellen dat gespecialiseerde Parkinson-verpleegkundigen wellicht het meest geschikt zijn om op te treden als persoonlijke casemanager. Ten tweede kunnen videobezoeken en zelfcontrole-apps helpen om enkele naden in de Parkinsonzorg te overbruggen. Ten derde zouden we de ontwikkeling van thuiszorgprogramma's voor mensen met de ZvP verwelkomen, in plaats van de meer ziekenhuisgebaseerde modellen van de huidige zorg.

Wat betreft het betalingsmodel voor Parkinsonzorg stellen we een experimentele brede UBM voor die financiële prikkels koppelt aan kwaliteitsindicatoren die relevant zijn voor mensen met de ZvP. 'Dichtheid' van zorgaanbiedersnetwerken zou in dit nieuwe model als kwaliteitsindicator kunnen fungeren, omdat een hoge dichtheid gunstige effecten heeft op de kwaliteit van zorg en de zorgkosten.

De sterkes en zwaktes van ons onderzoek in ogenschouw nemend, stellen we dat het toepassen van een mix van onderzoeksmethodologieën een grote kracht van dit proefschrift is. Andere sterke punten zijn de grote steekproefgroottes van onze analyses en de volledigheid van ons systematic review. Een beperking is het ontbreken van langetermijneffecten; d.w.z. langer dan vijf jaar na de diagnose. Een tweede beperking is dat onze beoordeling van UBM's niet ZvP-specifiek is. En tot slot hebben de activiteiten van ParkinsonNet mogelijk invloed gehad op de generaliseerbaarheid van onze bevindingen.

In onze toekomstvisie pleiten we voor meer onderzoek naar seksespecifieke doctor of patient delays om het 'eerdere' zorggebruik bij vrouwen met de ZvP te verklaren. We zouden graag een pilot zien waarin gespecialiseerde Parkinsonverpleegkundigen optreden als persoonlijke casemanagers voor mensen met de ZvP om te onderzoeken of dit de zorg voor mensen met de ZvP verbetert. We willen het gebruik van technologische innovaties zoals zelfcontrole-apps stimuleren. We zouden innovatieve zorgmodellen die zorg dichter bij huis brengen toejuichen. En tot slot pleiten we voor de ontwikkeling van een brede UBM voor ZVP-zorg met de dichtheid van zorgnetwerken als kwaliteitsindicator. We pleiten ook voor een pilot om de effecten van dit model op de kwaliteit van zorg en de zorgkosten te onderzoeken. Een betere, meer naadloze en betaalbarere zorgverlening voor mensen met de ZvP kan als blauwdrukvoorbeeld dienen voor andere chronische aandoeningen.

Resumo

Capítulo 1

A Doença de Parkinson (DP) é uma doença neurodegenerativa progressiva crónica, caracterizada por uma ampla gama de sintomas. Muitos prestadores de saúde diferentes estão envolvidos no tratamento de pacientes com DP, o que acarreta custos substanciais. Para atingir os melhores resultados, o atendimento aos pacientes com DP deve ser contínuo e sustentável. Nesta tese utilizamos as seguintes definições: *cuidado contínuo* é cuidado consistente e coerente, marcado por uma relação das partes ordenada, lógica e consistente, sem descontinuidades ou disparidades, uniformes em qualidade e combinados de forma imperceptível. *Cuidado sustentável* é o cuidado que pode ser sustentado ao longo do tempo, que é eficiente e não drena recursos finitos desnecessários.

Apesar dos esforços, o cuidado atual de pacientes com DP está frequentemente longe de ser contínuo e sustentável. Em primeiro lugar, os cuidados que os pacientes com DP recebem são bastante variáveis, o que implica que os cuidados não são "consistentes", "coerentes" ou "uniformes em qualidade". Em segundo lugar, os pacientes com DP não consideram os seus cuidados como "marcados por uma relação ordenada, lógica e consistente das partes". Terceiro, as questões financeiras nos sistemas de saúde impedem a prestação de cuidados contínuos e sustentáveis. E, finalmente, o atendimento aos pacientes com DP é prestado por profissionais de saúde dispersos, complicando a colaboração multidisciplinar e, como resultado, os profissionais de saúde tratam poucos pacientes com DP e não se tornam especialistas.

O objetivo desta tese é estudar medidas potenciais para alcançar um cuidado contínuo e sustentável de pacientes com DP. Isso resulta nas seguintes questões de pesquisa: Como é a percurso atual do paciente com DP? Quais são as descontinuidades atuais no atendimento aos pacientes com DP? Os modelos de pagamento baseados em resultados podem contribuir para um cuidado contínuo e sustentável de pacientes com DP? O cuidado de pacientes com DP será mais contínuo e sustentável se os prestadores partilharem mais pacientes?

Capítulo 2

No Capítulo 2, usamos o banco de dados nacional de reclamações médicas holandês para reconstruir o percurso de pacientes com PD por sexo durante os primeiros cinco anos após o diagnóstico. Numa análise tempo-para-evento usando dados dos seguros de saúde de todos os pacientes com diagnóstico de DP entre 2012 e 2016 nos Países Baixos, identificamos os momentos em que esses pacientes receberam cuidados de neurologistas, terapeutas de saúde aliados e médicos de clínica geral. Da mesma forma, identificamos os momentos em que marcos clínicos relevantes foram alcançados: a ocorrência de complicações relacionadas com a DP (por exemplo, pneumonia, lesão ortopédica e hospitalização devido à DP), internamento em lares de idosos e óbito.

A análise incluiu dados de 13.518 homens e 8.775 mulheres com DP recém diagnosticada. Afigura-se que, após o diagnóstico, as mulheres consultaram o seu clínico geral em média 10 dias e o seu fisioterapeuta em média 89 dias mais cedo do que os homens. Quanto a consultas a neurologistas, a diferença entre homens e mulheres foi pouca. Aproximadamente dois anos após o diagnóstico, ocorre a primeira complicação relacionada com a DP (mulheres: 1,8 anos; homens: 2,3 anos). Em cinco anos, 37,9% das mulheres haviam consultado pelo menos uma vez um terapeuta ocupacional e 18,5% um terapeuta da fala. Os números correspondentes para os homens foram 33,1% e 23,7%. Além disso, 72,9% das mulheres e 68,7% dos homens experimentaram pelo menos uma complicação relacionada com a DP; 27,5% das mulheres e 22,5% dos homens foram internados num lar de idosos. No período de cinco anos após o diagnóstico, 14,6% das mulheres e 18,3% dos homens faleceram. Esta diferença entre sexos pode ser devido a um atraso do médico ou paciente no processo de diagnóstico em mulheres, e não devido a uma progressão mais agressiva da doença nas mulheres.

Capítulo 3

No Capítulo 3, identificamos as principais necessidades dos pacientes com DP usando a abordagem Voz do Cliente (VoC): esta é uma nova metodologia desenvolvida originalmente no campo da indústria para sondar as necessidades dos clientes. Um grupo de 12 entrevistadores realizou entrevistas a pacientes com DP (n = 20), parentes (n = 12) e profissionais de saúde (n = 11). Os entrevistadores identificaram dez necessidades frequentemente mencionadas e combinaram as citações mais informativas num vídeo abrangente, que foi mostrado aos entrevistados numa reunião de consenso. Nessa reunião, os entrevistados prioritizaram as dez necessidades identificadas, atribuindo no máximo três pontos a cada uma das diferentes necessidades. Era possível alocar todos os três pontos a uma única necessidade ou dividi-los entre duas ou três necessidades. O número total de pontos representa a importância do tema.

A abordagem de VoC revelou que as necessidades mais urgentes do paciente diziam respeito ao domínio social, emocional ou doméstico, e não aos aspectos biomédicos da doença. As principais necessidades não atendidas foram: (1) maior autogestão; (2) melhor colaboração interdisciplinar entre diferentes prestadores de saúde; (3) mais tempo para discutir os cenários futuros e possíveis; e (4) um prestador de saúde atuando como gerente de caso pessoal, seja para resolver problemas diretamente ou para direcionar os pacientes com DP ao profissional mais bem equipado para resolver o problema em questão. Esses resultados podem servir para otimizar o atendimento aos pacientes com DP.

Capítulo 4

O Capítulo 4 descreve uma revisão dos efeitos dos modelos de pagamento baseados em resultados (OBPMs) na qualidade do atendimento e nos custos de saúde. Em primeiro lugar, desenvolvemos uma definição para OBPMs: um modelo de pagamento em saúde em que os pagamentos de incentivos relacionados ao desempenho dos prestadores de saúde dependem em pelo menos 10% dos resultados do atendimento prestado, sendo especialmente pensado para estimular efeitos favoráveis em termos de qualidade de cuidados ou custos de saúde. Em seguida, pesquisamos quatro fontes de dados para identificar os modelos: 1) bancos de dados de literatura científica; 2) sites de agências governamentais e científicas relevantes; 3) listas de referências dos artigos incluídos; 4) especialistas na área. Selecionamos apenas estudos que examinaram o impacto do modelo de pagamento na qualidade e / ou custos. Uma síntese de evidências foi usada para vincular características específicas aos efeitos sobre a qualidade do atendimento ou custos de saúde.

Incluímos 88 artigos, descrevendo 12 OBPMs. Nenhum deles focava especificamente a DP, embora todos os modelos tenham como alvo populações com condições crónicas. Distinguem-se dois grupos de OBPMs quanto à suas especificidades: OBPMs restritos, que contêm apenas incentivos financeiros explícitos para um desempenho de qualidade medido objetivamente; e OBPMs amplos, que combinam orçamentos globais e partilha de riscos para grupos de fornecedores multidisciplinares com incentivos financeiros explícitos para qualidade. A maioria (5 dos 9) OBPMs restritos mostrou efeitos positivos na qualidade; os outros tiveram efeitos mistos (2) ou negativos (2). Os efeitos dos OBPMs restritos sobre a utilização ou custos de saúde, no entanto, foram desfavoráveis (3) ou desconhecidos (6). Os três OBPMs amplos mostraram todos os efeitos positivos na qualidade do atendimento, ao mesmo tempo em que reduziram o crescimento dos custos de saúde. Embora apenas três OBPMs amplos puderam ser incluídos nesta análise, estes foram os que tiveram efeitos mais favoráveis na qualidade do atendimento e na utilização/custo dos cuidados de saúde.

Capítulo 5

No capítulo 5, observamos que os pacientes com DP criam as chamadas "redes de partilha de pacientes" de prestadores de saúde ao escolher os seus próprios prestadores de saúde. Essas redes diferem em "densidade": o número de pacientes que os prestadores partilham. Quisemos descobrir se redes mais densas de prestadores de serviços de saúde estavam relacionadas com melhores resultados para os pacientes, menor utilização de serviços de saúde e menores custos de saúde de DP. Usámos dados de seguros de saúde de todos os pacientes com DP nos Países Baixos entre 2012 e 2016. Incluímos todas as consultas a prestadores de saúde mais frequentemente envolvidos no atendimento a pacientes com DP. Para avaliar a densidade das redes de prestadores de saúde, usámos o número médio de pacientes que os prestadores de saúde partilham como um proxy para a densidade da sua colaboração (pontuação de densidade). Calculámos a pontuação de densidade média por paciente com DP e por população hospitalar. Visualizámos gráficos de colaboração dos três hospitais com pontuações mais altas e mais baixas, utilizando t-distributed Stochastic Neighbour Embedding (t-SNE). Usando análises de regressão logística e linear, estimámos a relação entre densidade e resultados de saúde, utilização de saúde e custos de saúde.

A pontuação de densidade média variou consideravelmente por paciente (média 6,7; desvio padrão 8,2), bem como por hospital (média 3,9; desvio padrão 8,2). Ajustados para variáveis de confusão, as pontuações de densidade mais altas foram associadas a um menor risco de complicações relacionadas com DP e a menores custos de saúde, embora não com mortalidade. Pontuações de densidade mais altas foram associados a envolvimento mais frequente de neurologistas, fisioterapeutas e terapeutas ocupacionais, mas a um envolvimento menos frequente de psicólogos. Portanto, propomos o uso de pontuações de densidade como indicador de qualidade em redes de prestadores de saúde.

Capítulo 6

No Capítulo 6, discutimos como poderíamos melhorar o atendimento de pacientes com DP, transferindo-o para a casa do paciente. O cuidado de pacientes com DP é frequentemente mal planeado: pacientes com DP que estão hospitalizados frequentemente sofrem atrasos no tratamento, medicamentos contraindicados, imobilidade prolongada, estadias prolongadas e alta mortalidade. Apesar dos benefícios documentados de receber cuidados de médicos com experiência em DP, muitos pacientes com DP não recebem esses cuidados especializados. Além disso, os modelos de cuidado atuais frequentemente exigem que os indivíduos mais velhos com mobilidade, cognição e capacidade de condução de veículos prejudicadas sejam transportados por cuidadores informais para grandes centros médicos urbanos.

Mover o atendimento para a casa do paciente tornaria o atendimento de DP mais centrado no paciente. Fatores demográficos, incluindo o envelhecimento da população, e fatores sociais, como a fragmentação familiar, aumentarão a necessidade de cuidados domiciliares. Os avanços tecnológicos, principalmente a capacidade de avaliar e prestar cuidados remotamente, possibilitarão a transição do cuidado para o domicílio. No entanto, apesar de ser promissora, esta próxima geração de atendimento domiciliar terá que superar barreiras, incluindo modelos de seguro desatualizados e uma divisão tecnológica. Uma vez iniciado, o atendimento domiciliar aumentará o acesso a atendimento de alta qualidade para o número crescente de pacientes com DP.

Capítulo 7

No Capítulo 7, interpretamos as nossas constatações e formulamos conclusões. Começamos com uma visão geral das principais descobertas. Primeiro, a reconstrução do percurso do paciente com Parkinson durante os primeiros cinco anos após o diagnóstico revelou profundas diferenças entre sexos: as mulheres consultam a maioria dos profissionais de saúde antes do diagnóstico e com mais frequência do que os homens (Capítulo 2). Em segundo lugar, as principais necessidades não atendidas dos pacientes são: : (1) maior autogestão; (2) melhor colaboração interdisciplinar entre diferentes prestadores de saúde; (3) mais tempo para discutir os cenários futuros e possíveis; e (4) um prestador de saúde atuando como gerente de caso pessoal (Capítulo 3). Terceiro, OBPMs amplos parecem ter os efeitos mais promissores quanto à qualidade do atendimento e custos de saúde (Capítulo 4). Quarto, redes mais densas de prestadores de saúde estão associadas a menor ocorrência de complicações relacionadas com DP, maior utilização de saúde, mas menores custos (Capítulo 5). Finalmente, os cuidados devem ser oferecidos mais perto da casa do paciente (Capítulo 6).

As nossas interpretações são as seguintes. O cuidado de pacientes com DP nos Países Baixos não é nem contínuo nem sustentável. Todas as descontinuidades identificadas dizem respeito a aspectos organizacionais, e não a aspectos biomédicos. A descontinuidade identificada de diferenças inexplicáveis de sexo no uso de cuidados de saúde pode ser devido ao atraso do médico ou do paciente no processo de diagnóstico em mulheres. As descontinuidades mais urgentes relatadas pelos próprios pacientes com DP dizem respeito ao domínio social, emocional ou doméstico. Para identificar descontinuidades no atendimento, a perspectiva dos pacientes com DP deve ser incluída, e o método Voz do Cliente pode auxiliar.

Oferecemos algumas sugestões sobre como lidar com essas descontinuidades. Primeiro, a forma como os prestadores estão organizados e dividem tarefas deve ser repensada. Argumentamos que enfermeiras especializadas em Parkinson são as mais adequadas para atuar como gerentes de casos pessoais. Em segundo lugar, as visitas de vídeo e os aplicativos de automonitorização podem ajudar a superar algumas falhas no tratamento de DP. Terceiro, gostaríamos de desenvolver programas de cuidados domiciliares para DP, em vez dos modelos de cuidados atuais mais baseados em hospitais.

Com relação ao modelo de pagamento para atendimento em DP, sugerimos um OBPM amplo experimental que vincule incentivos financeiros a indicadores de qualidade relevantes para DP. A 'densidade' poderia, portanto, atuar como um indicador de qualidade neste novo modelo, em que uma densidade alta teria efeitos favoráveis na qualidade dos cuidados e nos custos de saúde.

Resumindo os pontos fortes e as limitações da pesquisa, parece-nos que o uso de uma combinação de metodologias de pesquisa é um ponto forte importante. Outros pontos fortes são as grandes amostras de nossas análises e a abrangência da nossa revisão sistemática. Uma limitação é a não inclusão de efeitos de longo prazo; ou seja, além dos cinco primeiros anos após o diagnóstico. Uma segunda limitação é que nossa revisão sobre OBPMs, que não é específica para PD. E, finalmente, as atividades do ParkinsonNet podem ter influenciado a generalização das nossas descobertas em vários capítulos.

Quanto ao futuro, defendemos mais pesquisas sobre atrasos de médicos ou pacientes específicos por sexo em DP para explicar o uso "precoce" de vários serviços de saúde em mulheres com DP. Recomendamos um teste-piloto em que enfermeiras especializadas em DP atuem como gerente de caso pessoal para pacientes com DP, para confirmar se isso pode tornar o cuidado aos pacientes com DP mais contínuo. Gostaríamos de otimizar o uso de inovações tecnológicas, como aplicativos de automonitorização. Sugerimos modelos de cuidado inovadores que atendam pacientes com DP mais perto de casa. E, por fim, defendemos o desenvolvimento de um OBPM para o atendimento de DP com densidade como indicador de qualidade incorporado. Incentivamos um teste-piloto para estudar os efeitos desse modelo na qualidade do atendimento e nos custos de saúde. Uma prestação de cuidados de DP melhor, mais contínua e mais sustentável pode servir como um exemplo de modelo para outras condições crónicas.

Dankwoord

Beste Bas en Patrick, ik ben dankbaar dat ik twee van zulke kanjers als promotoren heb mogen hebben. Ik heb van jullie veel geleerd en koester grote bewondering voor jullie kwaliteiten. Bas, jouw vermogen om mensen te motiveren en te inspireren lijkt grenzeloos. Patrick, ik bewonder jouw bijzonder fijne (politieke) gevoel en analytische vaardigheden.

Beste Marjan en Marit, dank voor de begeleiding als co-promotoren. Een promotietraject gaat onontkoombaar gepaard met ups en downs. Zeker in de periodes dat ik het lastig had (het mislopen van opleidingsplekken; het drie maanden zonder contract zitten) heb ik me door jullie altijd goed gesteund gevoeld.

Geachte leden van de manuscriptcommissie, professor Assendelft, professor Rovers, doctor van Vugt. Dank voor het lezen van de van mijn manuscript en jullie aanwezigheid tijdens de promotie ceremonie.

Geachte leden van de oppositie, professor Minkman, professor Oertelt, doctor Westra. Dank voor het zitting nemen als opponent in de promotiecommissie.

Beste Henry, ook jou dank ik voor je deelname aan de oppositie. Maar ik dank je ook voor je altijd constructieve kritische houding, die mij een betere dokter maakt. Wat een voorrecht om met jou als opleider te mogen beginnen aan de (beste) neurologie opleiding.

Beste medeauteurs, in het bijzonder Marten, Stef en Femke. Dank voor jullie aandeel in de verschillende hoofdstukken van dit proefschrift, en jullie inzet om de onderzoeken en de artikelen steeds beter te maken.

Geachte professor Schut, geachte doctor Eijkenaar, dank voor jullie inzet en uitgebreide inbreng bij Hoofdstuk 4.

Beste Jesse, speciale dank voor jouw enorme hulp bij de statistische analyses van Hoofdstukken 2 en 5. En ook dank voor je eeuwige geduld bij het mij (herhaaldelijk) uitleggen van mijn soms wel erg basale vragen.

Beste Kees, Jij hebt letterlijk de laatste hand (je eigen!) aan mijn proefschrift gelegd. Wat leuk dat jij mijn kaft wilde ontwerpen, en dank voor je inspanningen!

Beste Michel en Kyah, dank voor al jullie hulp bij het drukproces.

Beste kelderbewoners, in het bijzonder Joost, Niek, Wieteke, Inger, Florien, Yvonne, en Karel-Peter, wat denk ik met veel plezier terug aan de jaren “in de catacomben”. Met een grote glimlach denk ik terug op onze politieke twisten en discussies met bijbehorende steken onder water. Wat was het een genoegen om daarbinnen dan weer als rode lap, dan weer als stier te dienen. Heerlijk. Ik denk ook aan andere extra-curriculaire zaken zoals de voetbalpoultjes en de reply-all mailtjes. Toch denk ik niet in de laatste plaats aan de inhoudelijke discussies en voordrachten binnen het toenmalige Celsus die we allen met onze verschillende achtergronden van veel kleur konden voorzien! Dat Celsus was een goed groepje!

Beste (oud-)collegae van ParkinsonNet. Zelden heb ik deel mogen uitmaken van zo’n geïnspireerde groep; een zeer multidisciplinaire groep, tot het bot gemotiveerd voor dat ene doel: het leven van mensen met Parkinson te verbeteren. Het was een leuke en zeer motiverende omgeving om in te werken. Ik denk in het bijzonder met plezier terug op de grote congressen die we jaarlijks organiseerde. Een feest om aan bij te mogen dragen.

Lieve (schoon)familie en vrienden, in het bijzonder Jet, Patrick, Frits, Armando, Manuela, Willem, Tim, Marta, João, Tim, Maaike, Jelle, Liz, Kamil, en Renée. Dank voor alle steun en warmte de afgelopen jaren. Door mijn dubbele banen heb ik minder tijd met jullie kunnen doorbrengen dan gewenst, want om te leven in jullie aanwezigheid is heerlijk. Waarom zitten er maar 24 uren in een dag?

Lieve pappa en mamma, dankzij jullie heb ik me kunnen ontwikkelen tot de persoon die ik ben, en heb ik het leven dat ik leef. Ik kan jullie daar nooit genoeg voor danken. Ook mijn dank voor alle hulp met de kinderen en tijdens mijn dienstweken. Jullie staan echt altijd voor ons klaar.

Lieve Joana, jij bent de rots in mijn leven. Je hebt het hele traject met me meegemaakt. Ook als ik het even niet meer zag zitten, wist jij mij altijd te motiveren. Dank voor je onvoorwaardelijke steun en liefde. Ondertussen zijn we ook getrouwd, ben je in Nederland komen wonen en hebben we twee fantastische kinderen. Je vult me aan, verbetert mijn zwakheden en laat mijn sterke kanten floreren. Ik ben een beter mens sinds ik je ken en zonder jou was dit proefschrift ook nooit gelukt.

Lieve Frederik en Henriëtte, ik kan hier moeilijk opschrijven dat het proefschrift er dankzij jullie is gekomen (soms eerder: ondanks), maar wat ben ik blij dat ik jullie in mijn leven heb: twee prachtige kinderen die zo lief voor elkaar zijn. Wat ben ik trots op jullie! Jullie ontwikkelen je fantastisch en doen me beseffen wat een rijk leven ik heb.

About the author

Floris Pieter Vlaanderen was born on 23rd of October 1987 in Amsterdam. He grew up in Muiderberg and attended Sint Vituscollege in Bussum. In 2006, he moved to Rotterdam to start medical school at the Erasmus University Medical Center. Having developed an interest in healthcare politics, Floris enrolled in a Bachelor's Degree in Policy and Management in Healthcare (PMH) at the Erasmus University Rotterdam. This degree gave him the opportunity to do an internship at the Dutch parliament in the Hague, where he worked for 6 months. Floris attended both degrees simultaneously, having obtained his BSc in PMH in 2011 and his MSc in Medicine in 2013.

After finishing his degrees, Floris started working as a medical doctor at the neurology department of the Flevoziekenhuis in Almere. In 2014, Floris got the opportunity to start his PhD in the field of Parkinson's disease at Radboudumc Nijmegen. This research program allowed him to combine his interests in neurology and the organisation of healthcare; this thesis is the result of his work. He combined this research work with clinical positions at the neurology departments of the Radboudumc (2015), Gelre Ziekenhuis Apeldoorn (2019), Amsterdam University Medical Center location AMC (2019) and OLVG Hospital Amsterdam (2020-current). He will start his specialisation in neurology in his current hospital on the 1st of January 2022.

Floris lives with his wife Joana, his son Frederik and his daughter Henriëtte in Muiderberg. In his free time, he plays contract bridge; he represented The Netherlands in various international under-25 tournaments of this sport.

Dissertations of the disorders of movement research group, Nijmegen

Parkinson Center Nijmegen (ParC)

- Jasper E. Visser. The basal ganglia and postural control. Radboud University Nijmegen, June 17th 2008.
- Maaike Bakker. Supraspinal control of walking: lessons from motor imagery. Radboud University Nijmegen, May 27th 2009.
- W. Farid Abdo. Parkinsonism: possible solutions to a diagnostic challenge. Radboud University Nijmegen, October 7th 2009.
- Samyra H.J. Keus. Physiotherapy in Parkinson's disease. Towards evidence-based practice. Leiden University, April 29th 2010.
- Lars B. Oude Nijhuis. Modulation of human balance reactions. Radboud University Nijmegen, November 29th 2010.
- Maarten J. Nijkrake. Improving the quality of allied health care in Parkinson's disease through community-based networks: the ParkinsonNet health care concept. Radboud University Nijmegen, November 29th 2010.
- Rick C.G. Helmich. Cerebral reorganization in Parkinson's disease. Radboud University Nijmegen, May 24th 2011.
- I.B. Bruinsma. Amyloidogenic proteins in Alzheimer's and Parkinson's disease. Interaction with chaperones and inflammation. Radboud University Nijmegen, September21st 2011.
- Charlotte A. Haaxma. New perspectives on preclinical and early stage Parkinson's disease. Radboud University Nijmegen, December 6th 2011.
- Johanna G. Kalf. Drooling and dysphagia in Parkinson's disease. Radboud University Nijmegen, December 22nd 2011.
- Anke H. Snijders. Tackling freezing of gait in Parkinson's disease. Radboud University Nijmegen, June 4th 2012.
- Bart F.L. van Nuenen. Cerebral reorganization in premotor parkinsonism. Radboud University Nijmegen, November 22nd 2012.
- Wandana Nanhoe-Mahabier. Freezing of physical activity in Parkinson's disease, the challenge to change behavior. Radboud University Nijmegen, February 13th 2013.
- Marlies van Nimwegen. Promotion of physical activity in Parkinson's disease, the challenge to change behavior. Radboud University Nijmegen, March 6th 2013.

- Arlène D. Speelman. Promotion of physical activity in Parkinson's disease, feasibility and effectiveness. Radboud University Nijmegen, March 6th 2013.
- Tjitske Boonstra. The contribution of each leg to bipedal balance control. University Twente, June 6th 2013.
- Marjolein A van der Marck. The Many faces of Parkinson's disease: towards a multifaceted approach? Radboud University Nijmegen, January 20th 2014.
- Katrijn Smulders. Cognitive control of gait and balance in patients with chronic stroke and Parkinson's disease. Radboud University Nijmegen, May 21st 2014.
- Marjolein B. Aerts. Improving diagnostic accuracy in parkinsonism. Radboud University Nijmegen, June 27th 2014.
- Maartje Louter. Sleep in Parkinson's disease. A focus on nocturnal movements. Radboud University Nijmegen, February 13th 2015.
- Frederick Anton Meijer. Clinical Application of Brain MRI in Parkinsonism: From Basic to Advanced Imaging, Radboud University Nijmegen, June 23th 2015.
- Jorik Nonnekes. Balance and gait in neurodegenerative disease: what startle tells us about motor control, Radboud University Nijmegen, September 25th 2015.
- Martijn van der Eijk. Patient-centered care in Parkinson's disease. Radboud University Nijmegen, December 1st 2015.
- Ingrid Sturkenboom. Occupational therapy for people with Parkinson's disease: towards evidence-informed care. Radboud University Nijmegen, February 11th 2016.
- Merel M. van Gilst. Sleep benefit in Parkinson's disease. Radboud University Nijmegen, April 13th 2016.
- Arno M. Janssen. Transcranial magnetic stimulation - measuring and modeling in health and disease. Radboud University Nijmegen, June 2nd 2016.
- Annette Plouvier. De ziekte van Parkinson, een gezamenlijke reis van huisarts en patiënt. Radboud University Nijmegen, juni 15th 2017.
- Nico Weerkamp. Parkinson's disease in long-term-care facilities. Radboud University Nijmegen, September 1st 2017.
- Digna de Kam. Postural instability in people with chronic stroke and Parkinson's disease: dynamic perspectives Radboud University Nijmegen, October 4th 2017.
- Freek Nieuwhof. The complexity of walking: Cognitive control of gait in aging and Parkinson's disease Radboud University Nijmegen, October 27th 2017.
- Koen Klemann. A molecular window into Parkinson's disease. Radboud University Nijmegen, November 3th 2017.
- Claudia Barthel. Moving beyond: freezing of gait in Parkinson's disease. Radboud University Nijmegen, April 4th 2018.
- Esther Bekkers. Freezing and postural control in Parkinson's disease. Defense at KU Leuven, May 15th 2018.
- Erik te Woerd. Feeling the beat: The neurophysiology of cueing in Parkinson's disease. Radboud University Nijmegen, January 18th 2019.
- Ana L. Silva de Lima. Quantifying Parkinson's disease: the use of technology for objective assessment of motor symptoms. Radboud University Nijmegen, March 26th 2019.
- Anna Santaella Tortós-Sala. Tackling Parkinson's disease: a proteomic approach to biomarkers and regenerative therapy. Radboud University Nijmegen, October 22nd 2020.

Non-Parkinsonian disorders of movement

- Sacha Vermeer. Clinical and genetic characterization of autosomal recessive cerebellarataxias. Radboud University Nijmegen, April 5th 2012.
- Susanne T. de Bot. Hereditary spastic paraplegias in the Netherlands. Radboud University Nijmegen, December 20th 2013.
- Catherine C.S. Delnooz. Unraveling primary focal dystonia. A treatment update and new pathophysiological insights. Radboud University Nijmegen, January 7th 2014.
- Ella M.R. Fonteyn. Falls, physiotherapy, and training in patients with degenerative ataxias. Radboud University Nijmegen, June 29th 2016.
- B.S. Hoffland. Investigating the role of the cerebellum in idiopathic focal dystonia. Radboud University Nijmegen, March 22nd 2017.

Vascular disorders of movement – The Radboud Stroke centre

- Liselore Snaphaan. Epidemiology of post stroke behavioral consequences. Radboud University Nijmegen, March 12th 2010.
- D. de Jong. Anti-inflammatory therapy and cerebrospinal fluid diagnosis in Alzheimer's disease. Radboud University Nijmegen, September 21st 2010

- N.M. Timmer. The interaction of heparin sulfate proteoglycans with amyloid β protein. Radboud University Nijmegen, January 13th 2011.
- Karlijn F. de Laat. Motor performance in individuals with cerebral small vessel disease: an MRI study. Radboud University Nijmegen, November 29th 2011
- Anouk G.W. van Norden. Cognitive function in elderly individuals with cerebral small vessel disease. An MRI study. Radboud University Nijmegen, November 30th 2011.
- P.E. Spies. The reflection of Alzheimer disease in CSF. Radboud University Nijmegen, March 15th 2012.
- D. Slats. CSF biomarkers of Alzheimer's disease; serial sampling analysis and the study of circadian rhythmicity. Radboud University Nijmegen, September 21st 2012.
- Rob Gons. Vascular risk factors in cerebral small vessel disease. A diffusion tensor imaging study. Radboud University Nijmegen, December 10th 2012.
- Loes C.A. Rutten-Jacobs. Long-term prognosis after stroke in young adults. Radboud University Nijmegen, April 14th 2014
- M.K. Herbert. Facing uncertain diagnosis. The use of CSF biomarkers for the differential diagnosis of neurodegenerative disease. Radboud University Nijmegen July 8th 2014.
- Noortje A.M.M. Maaijwee. Long-term neuropsychological and social consequences after stroke in young adults. Radboud University Nijmegen, June 12th 2015.
- M. Müller. Footprints of Alzheimer's disease. Exploring proteins and microRNAs as biomarkers for differential diagnosis. Radboud University Nijmegen, April 18th 2016.
- K.A. Bruggink. Amyloid-B and amyloid associated proteins in the pathology and diagnosis of Alzheimer's Disease. Radboud University Nijmegen, April 25th 2016.
- Anil M. Tuladhar. The disconnected brain: mechanisms of clinical symptoms in small vessel disease. Radboud University Nijmegen, October 4th 2016.
- Pauline Schaapsmeerders. Long-term cognitive impairment after first-ever ischemic stroke in young adults: a neuroimaging study. Radboud University Nijmegen, January 24th 2017.
- Inge W.M. Van Uden. Behavioral consequences of cerebral small vessel disease. An MRI approach. Radboud University Nijmegen, February 14th 2017.
- Renate Arntz. Long-term risk of vascular disease and epilepsy after stroke in young adults. Radboud University Nijmegen, February 16th 2017.
- Helena Maria Van Der Holst. Mind the step in cerebral small vessel disease. Brain changes in motor performance. Radboud University Nijmegen, April 5th 2017.
- E.M.C. van Leijsen. Unraveling the heterogeneity of cerebral small vessel disease; from local to remote effects. Radboud University Nijmegen, November 19th 2018.
- S.J. Ooms. Sleep well, age well. Assessing sleep disruption as a player in Alzheimer's disease. Radboud University Nijmegen, November 30th 2018.
- Linda J.C. van Waalwijk van Doorn. Cerebrospinal fluid biomarker assays for Alzheimer's disease: standardization, validation and analysis of confounders. Radboudumc, Nijmegen, August 27th 2020.

Neuromuscular disorders of movement

- Mireille van Beekvelt. Quantitative near infrared spectroscopy (NIRS) in human skeletal muscle. Radboud University Nijmegen, April 24th 2002.
- Johan Hiel. Ataxia telangiectasia and Nijmegen Breakage syndrome, neurological, immunological and genetic aspects. Radboud University Nijmegen, April 23th 2004.
- Gerald JD Hengstman. Myositis specific autoantibodies, specificity and clinical applications. Radboud University Nijmegen, September 21st 2005.
- M. Schillings. Fatigue in neuromuscular disorders and chronic fatigue syndrome, a neurophysiological approach. Radboud University Nijmegen, November 23th 2005.
- Bert de Swart. Speech therapy in patients with neuromuscular disorders and Parkinson's disease. Diagnosis and treatment of dysarthria and dysphagia. Radboud University Nijmegen, March 24th 2006.
- J. Kalkman. From prevalence to predictors of fatigue in neuromuscular disorders. The building of a model. Radboud University Nijmegen, October 31st 2006.
- Nens van Alfen. Neuralgicamyotrophy. Radboud University Nijmegen, November 1st 2006.
- Gea Drost. High-density surface EMG, pathophysiological insights and clinical applications. Radboud University Nijmegen, March 9th 2007.

- Maria Helena van der Linden. Pertubations of gait and balance: a new experimental setup applied to patients with CMT type 1a. Radboud University Nijmegen, October 6th 2009.

- Jeroen Trip. Redefining the non-dystrophic myotonic syndromes. Radboud University Nijmegen, January 22nd 2010.

- Corinne G.C. Horlings. A weak balance: balance and falls in patients with neuromuscular disorders. Radboud University Nijmegen, April 1st 2010.

- Edith Cup. Occupational therapy, physical therapy and speech therapy for persons with neuromuscular diseases, an evidence based orientation. Radboud University Nijmegen, July 5th 2011.

- Alide Tieleman. Myotonic dystrophy type 2, a newly diagnosed disease in the Netherlands. Radboud University Nijmegen, July 15th 2011.

- Nicol Voermans. Neuromuscular features of Ehlers-Danlos syndrome and Marfan syndrome. Radboud University Nijmegen, September 2nd 2011.

- Allan Pieterse. Referral and indication for occupational therapy, physical therapy and speech- language therapy for persons with neuromuscular disorders. Radboud University Nijmegen, February 13th 2012.

- Bart Smits. Chronic Progressive External Ophthalmoplegia more than meets the eye. Radboud University Nijmegen, June 5th 2012.

- Ilse Arts. Muscle ultrasonography in ALS. Radboud University Nijmegen, October 31st 2012.

- M. Minis. Sustainability of work for persons with neuromuscular diseases. Radboud University Nijmegen, November 13th 2013.
- Willemijn Leen. Glucose transporter – 1 deficiency syndrome. Radboud University Nijmegen, June 26th 2014.

- Femke Seesing. Shared Medical appointments for neuromuscular patients and their partners. Radboud University Nijmegen, September 2nd 2016.

- Nicole Voet. Aerobic exercise and cognitive behavioral therapy in fascioscapulohumeral dystrophy: a model based approach. Radboud University Nijmegen , October 14th 2016.

- Barbara van der Sluijs. Oculopharyngeal muscular dystrophy (OPMD) in the Netherlands, beyond dysphagia and ptosis. Radboud University Nijmegen, December 11th 2017.

- Simone Knuijs. Prevalence of dysarthria and dysphagia in neuromuscular diseases and an assessment tool for dysarthria in adults. Radboud University Nijmegen, July 3th 2018.

- Marielle Wohlgemuth. A family based study of Facioscapulohumeral. Radboud University Nijmegen, November 7th 2018.

- Karlien Mul. The many faces of FSHD, opportunities and challenges on the road to therapies. Radboud University Nijmegen, 18 January 18th 2019.