Personal science in Parkinson's disease

A patient-led research study

Sara Riggare

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A patient-led research study

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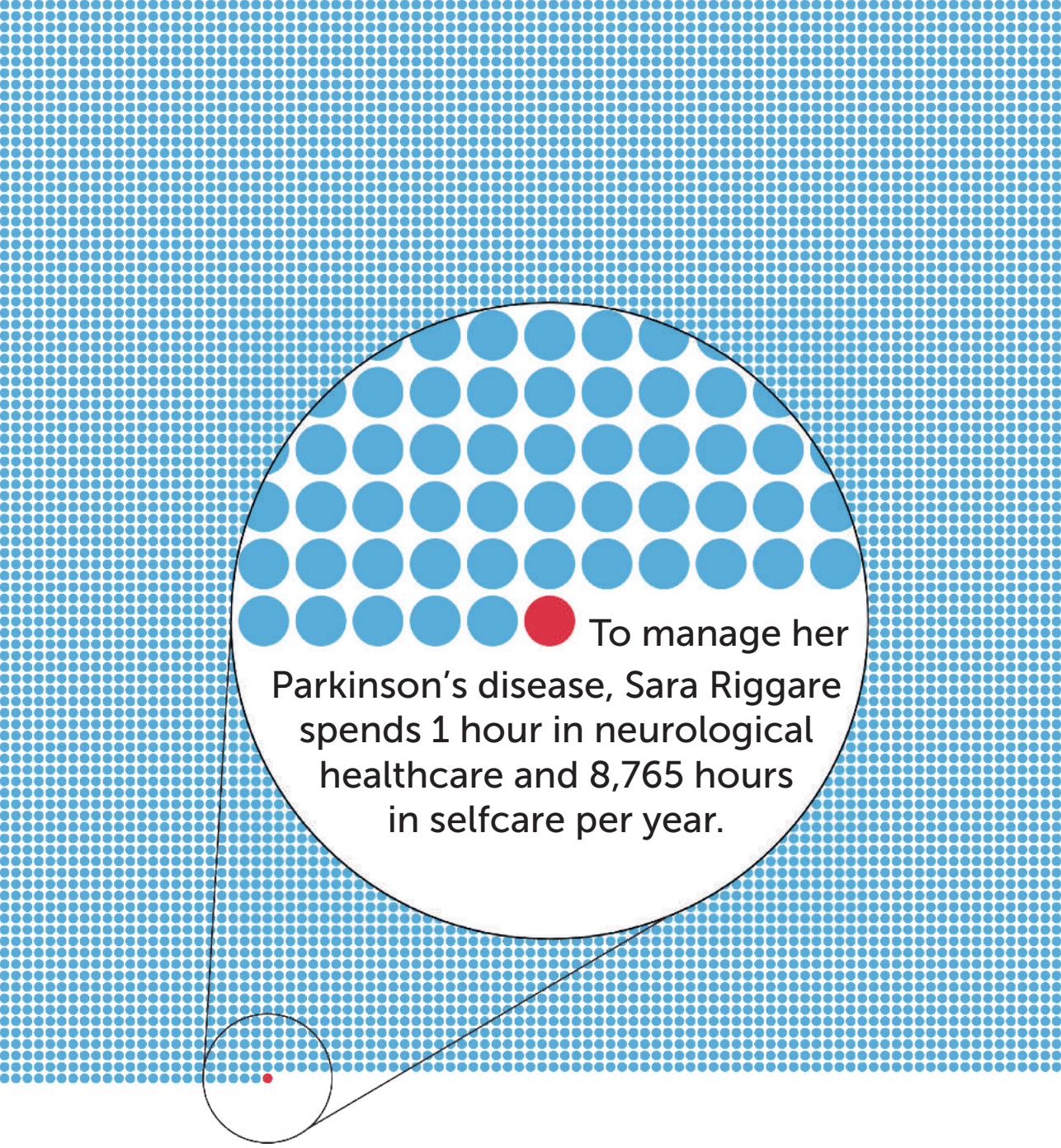

To manage her Parkinson's disease, Sara Riggare spends 1 hour in neurological healthcare and 8,765 hours in selfcare per year.

TABLE OF CONTENTS

PROLOGUE

"You have Parkinson's disease."

It was Thursday the 12th of June 2003. I was 32 years old, and I was walking through Stockholm on a warm summer's day. I had left my 4 months old baby with my mother-in-law for my doctor's appointment. Since my early teens, I had always known that my body didn't always respond the way I expected it to. A few years earlier, my neurologist had told me that I had dopa-responsive dystonia, a rare condition affecting the nerves' control of the muscles, resulting in tense muscles and reduced muscle control. Fortunately, the condition responded well to medication intended for Parkinson's disease, and a few years previously, I had started taking levodopa. The medication worked very well and made me able to move more freely than I had in a very long time. This was an important reason why we had decided to try to have a baby, the result of which was waiting for me at home.

The neurologist I saw that day, who was not the one I normally went to, was especially interested in dopa-responsive dystonia. I had booked the appointment because I wanted to know more about the current state of research in the field. I hadn't expected him to put me through the full neurological examination, which he did. The test included for example the finger-to-nose test, walking back and forth in the corridor and the pull test. As the meeting was drawing to a close, he said to me: "You don't have dopa-responsive dystonia, you have Parkinson's disease."

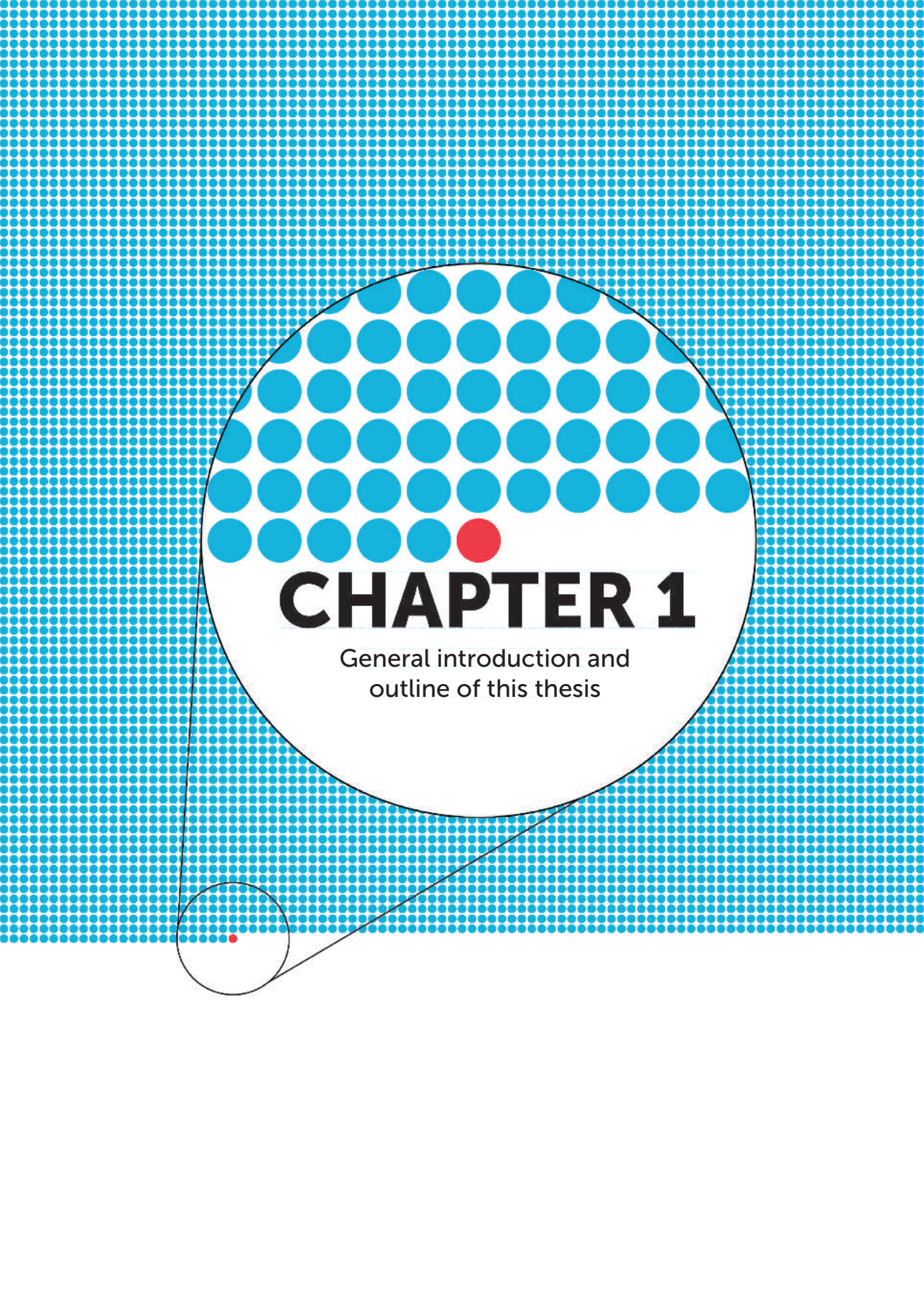
His words hit me like a ton of bricks, and I felt like I fell down into a black hole. How could I have Parkinson's disease? I was only 32, surely Parkinson's disease was something that only happened to old people? A thousand questions were bouncing around in my head but the shock made me unable to ask them. The neurologist stood up as a signal that our meeting was coming to an end. As he opened the door for me, he said: "Have a nice day."

This thesis is my attempt at giving others better tools than I had for making sense of and living with a life-changing diagnosis of a complex condition like Parkinson's disease.

GLOSSARY

Important terms and definitions	
Citizen science	active public involvement in scientific research
N-of-1 study/trial	"multiple crossover trials, usually randomized and often blinded, conducted in a single patient"[1]
Participatory science	synonym to citizen science
Patient-led research	in this thesis, patient-led research is defined as research led by patient researchers (see patient researchers)
Patient researcher	persons with lived experience of a disease, disability, or other health challenge who are openly using those experiences in doing research, within academia or in other contexts. This can also include a family member, partner, or similar of the person living with a disease.
Personal science	a framework of study for single subject research defined in this thesis as: *"the practice of exploring personally consequential questions by conducting self-directed N-of-1 studies using a structured empirical approach."* Complementary to conventional clinical research. In personal science the researcher and the person studied are one and the same. Can answer questions like "What works for me?" and includes self-tracking as a main method for data collection.
Quantified Self	a global social movement for self-trackers founded in San Francisco in 2008.
Selfcare	*"The ability of individuals, families and communities to promote health, prevent disease, maintain health, and cope with illness and disability with or without the support of a healthcare provider"*[2]. The absolute majority of research on selfcare is conducted by people without lived experience of the condition in question. In chronic diseases, patients typically become the primary caregiver, as they are the ones dealing 24/7 with the medical issues, which means that patients' selfcare is key for optimal health. For PD, selfcare includes, among other things, understanding of PD symptoms and available treatments (pharmacological and non-pharmacological), both on a group level (*What treatments can be expected to have an effect on PD?*) and on an individual level (*Which specific treatments have what effects on me?*).
Self-tracking	a method for data collection defined as "a process of deliberately collecting and structuring observations about one's own life". In this thesis, self-tracking is defined to be able to be used in personal science as well as in conventional clinical group research. Tools for self-tracking include different types of technology as well as pen and paper.

1 From: Kravitz R, Duan N (2014) *Design and Implementation of N-of-1 Trials: A User's Guide*.
2 From: World Health Organization (2019) *WHO consolidated guideline on self-care interventions for health: sexual and reproductive health and rights*.



CHAPTER 1

General introduction and outline of this thesis

GENERAL INTRODUCTION

This thesis seeks to enhance the understanding and development of selfcare in Parkinson's disease (PD) by exploring the use of **patient-led research** in the form of **personal science**. In this chapter, I will briefly describe PD, how the disease is managed clinically, outline some aspects of living with PD, and possibilities for selfcare of this condition. Thereafter, I will introduce the two concepts: that of *personal science* and that of *patient-led research*. Lastly in this chapter, I will describe the outline.

I have chosen to incorporate vignettes of my own experience of living with PD throughout this thesis, with the purpose of providing a fuller picture of the condition. The disruption it may cause to the reading flow is intentional and analogous to how living with PD disrupts the flow of everything I do.

Parkinson's disease

Parkinson's disease (PD) is a neurological condition first formally described by the London-based physician James Parkinson in 1817. In his book "An essay on the shaking palsy", Parkinson outlined the features of what he saw as an insufficiently characterised disease. His accounts were based on observations of in total six individuals, whom he saw in his clinic or in the streets of London [1]. Treatments that were used at the time, and which had limited or no effect, included bloodletting with leeches, stimulating fomentations and blisters [1]. More information about PD can be found in **Box 1.1** and information about current day clinical management of PD can be found in **Box 1.2**.

I see my neurologist once or twice a year, about half an hour every time. That's one hour per year in healthcare for my Parkinson's disease (PD). During the same year, I spend 8,765 hours in selfcare, applying my knowledge and experience together with what I get from my neurologist to manage a difficult condition to the best of my abilities. Of course, I don't mean that I spend all of the hours of every year actively managing my PD. I am however aware of the disease 24/7, also at night, since my reduced mobility often makes it difficult to turn over in bed.

Only during one single hour per year (the red circle in the image on the cover) am I in direct contact with neurological specialty care and its clinical practice and guidelines. And it's during this one hour per year that my condition is evaluated by my neurologist and when my treatment is prescribed. But it's during the 8,765 hours of selfcare (the many blue circles in the image on the cover) that I put my prescribed treatment into action. Because, let's face it, my doctor doesn't even know if I take my medications or not.

It is also during my 8,765 hours of selfcare that I can observe the effects of my treatment. And I do. Every day, some days every hour that I am awake, I observe and assess my physical and mental functionality. I am constantly vigilant, on the lookout for ways to help me answer questions such as: "When is it time for my next medication dose?", "Is my mobility good enough for a long walk?", "Will my energy last long enough to complete this task?"

Living with the constant variability and unpredictability of PD has made me a student of myself, and more importantly, turned me into a curious and studious patient researcher.

BOX 1.1: About Parkinson's disease.

About Parkinson's disease
Parkinson's disease (PD) is a neurodegenerative disease manifesting in numerous motor and non-motor symptoms. The condition is primarily diagnosed based on the presence of four cardinal symptoms: tremor, bradykinesia, rigidity, and gait problems [2–4].
The main breakthrough in PD treatment came with the introduction of levodopa in the late 1950s and early 1960s [5]. The effect on persons with PD was truly astonishing: *"Bedridden patients who were unable to sit up, patients who could not stand up from a sitting position, and patients who, when standing, could not start walking, performed all these activities after L-DOPA* with ease. They walked around with normal associated movements and they even could run and jump"* [6]. * L-DOPA = levodopa
The mean survival time after diagnosis in a group of patients treated in the period 1949-1964 has been reported to have been less than 11 years [7].
PD is the fastest growing neurological condition in the world [8]. In 2016, the disease affected approximately 6,1 million individuals worldwide, and this number is projected to double by the year 2030 [9].
Median age of onset is 60 years and the prevalence increases with age [2,3]
One common misconception is that PD exclusively affects the elderly. Between 5 and 10% of cases have young onset PD (YOPD), meaning their PD first manifested itself before the age of 50. For a small subset of that group, referred to as juvenile onset PD, disease onset occurred before 21 years of age [10].

Living with PD

Both the influence of PD and the effects of oral pharmacotherapy frequently have a very fluctuating nature, such that symptoms can differ immensely from day to day, from hour to hour, and even from one minute to the next [3,14]. Another related challenge is the need to balance medication effects and side effects, which may require careful titration of doses and timings [14]. When it comes to which symptoms persons with PD find most troublesome, studies find that the views are highly diverse and that quality of life is affected by both motor symptoms, non-motor symptoms, as well as the fluctuating response to medication including side effects [20–23]. Ultimately, what matters most to persons with PD is highly individual, and the treatment priorities and personalised goals in life will differ considerably from person to person.

BOX 1.2: Clinical management of PD.

Clinical management of PD
Since the introduction of levodopa, additional pharmaceutical therapies have been implemented to optimise the management of PD. These therapies have improved the possibilities to control both the motor and non-motor symptoms [2–4]. However, even with optional pharmacotherapy, PD significantly reduces quality of life, and increasingly so with longer disease duration [2–4,10,11].
Current primary treatment is oral pharmacotherapy, often with a complex and complicated regimen. There is a significant risk of side effects, particularly after longer disease duration [2–4,10–12]. The types, amounts, and combinations of drugs prescribed vary with, for example, national prescribing patterns [11], age of onset and duration of disease [12], and the use of concurrent brain surgery [13]. Typically the number of pills, and the number of times per day that this medication needs to be taken, increases with longer duration of the disease [14]. One study found the mean daily intake to be three pills for early stage PD and eight to nine pills for persons with PD with advanced stage disease [15].
One potential medication side effect is levodopa-induced dyskinesia (LID). LID manifests as abnormal involuntary movements with writhing and sometimes jerky movement patterns. LID can be mild and acceptable to the individual in the early phases of its development. However, LID can become very debilitating and socially disruptive for many [10,12,16,17]. The risk of LID is significantly higher for persons with YOPD and can be difficult to manage [10,12,16,17].
For persons with advanced disease, several device-aided therapies are available, and this includes brain surgery, such as Deep Brain Stimulation (DBS), and two types of infusion therapy, including intraduodenal levodopa pump therapy and apomorphine pump therapy [4].
Other essential elements of a successful management approach are physical and mental exercise, preferably supported by high-quality physiotherapy [2–4,18]. PD is best managed clinically by multidisciplinary healthcare teams [18,19]. Over 30 different professional disciplines can potentially add value to the lives of individuals affected by PD [2].

Living with PD includes an unavoidable focus on medication, without my medications I simply don't function. Now, in August 2021, my medical treatment for PD consists of five prescription drugs that are combined in different ways to be taken several times every day. With my neurologist as an occasional discussion partner, I have worked out a medication strategy that works for me. I try to take my pills at the same time every day. Of course, I don't always succeed, sometimes I oversleep and take the first dose late. If that happens, I usually adjust the rest of the doses of that day accordingly so that the time intervals between doses are the same as usual. Occasionally, I forget the time and miss the timing of a dose. If I miss a dose, even by as little as half-an-hour, the rest of my day is affected, at least up until the next dose, often longer. My movements will be even slower than usual, and my movement functionality will be even more unpredictable.

Despite considerable efforts over many decades, PD research has been unable to deliver a cure for the disease, and there is still no way to reliably slow down the progression of the disease [3,4,10–12]. Another unmet need is that there is still no

biomarker available to clearly and unambiguously evaluate the current state and/or the progression of the condition in individuals [3,23]. PD is considered by many to be one of the most complex conditions known to medicine, and this is definitely true for experts who have worked with individuals living with PD for a long time. It is certainly one of the most difficult conditions to manage, and these complexities only increase further with progressing disease. It is therefore very important for persons with PD to understand their medications, including for example doses, timings, and side effects [3,4,10–12,14,15,18,19]. And a particular challenge is the tremendous inter-individual variability in disease presentation, response to treatment (both in terms of efficacy and side effects), requirements for multidisciplinary care and personal preferences. This further emphasises that a highly individualised approach to both medical management and selfcare is essential.

Selfcare

Selfcare, or the "invisible work of being a patient" [24], is increasingly acknowledged as an important contributor to health, especially when it comes to chronic conditions such as PD [14,18,19,25–30]. Successfully managing PD includes, among other things, having an adequate level of knowledge about the disease itself, understanding the various medications and their side effects, being able to monitor symptoms, finding reliable sources of information, knowing when to take action, understanding the importance of lifestyle interventions such as exercise or diet, knowing how to navigate the healthcare system, and organising healthcare contacts and information [14,26,27,31–33]. More effective selfcare is associated with significant positive effects on health and healthcare utilisation [34–37].

This is especially important when considering that healthcare has been found to contribute to approximately 10% of our populations' overall health, whereas the remaining 90% can be attributed to individual factors, like genetics, social circumstances, and environmental exposure (50%), as well as to behavioural patterns (40%) [38]. Definitions for health and selfcare can be found in **Box 1.3**.

BOX 1.3: Health and selfcare.

Health and selfcare
In 2011, a new definition of health was suggested as: "the ability to adapt and to self-manage, in the face of social, physical and emotional challenges" [39].
In 2019, the World Health Organization published a report where selfcare was defined as "the ability of individuals, families and communities to promote health, prevent disease, maintain health, and cope with illness and disability with or without the support of a healthcare provider" [40].

Living with a complex disease, like PD, means there's a lot of extra work to do, and a lot of time goes into managing and organising my pills. My first dose is due at 7 am so regardless of whether this is a working day, a weekend, or a holiday, my alarm is set. I keep a pill organiser with one compartment for each day of the week next to my bed together with a bottle of water. When the alarm goes off, I find my pill organiser, open the compartment corresponding to the current day of the week and tip the pills into my left hand. I then check them using my right hand, making sure that they are the right kinds and the correct number before I put them in my mouth and swallow them down with water. I am very careful not to drop any pills, the pill organiser, or the water bottle, which can be easier said than done, since I am very clumsy and slow when it has been long since my previous dose. I also try to be as quiet as possible as to, if possible, avoid waking up my husband. Most of the time I then try to get back to sleep for 30 minutes or so to give the meds a chance to "kick in" before I rise and start my day. The remaining six doses are taken throughout the day, at the scheduled time or slightly later. To avoid unnecessary side effects, for example the involuntary movements of levodopa-induced dyskinesia (LID), I don't want to take my pills too closely together. Despite all my efforts, I sometimes do experience LID.

Patients in general see selfcare as a means to living their life to the fullest, in the presence of health challenges [41,42]. In patients' view, selfcare includes, among other things, managing your treatments, lifestyle factors, balancing activity and recuperation, collecting and organising information as well as navigating and coordinating healthcare contacts [32,42–44]. Clinicians' view of selfcare has been reported to be less holistic, and limited to activities relating to the healthcare context [41]. In fact, some studies found that healthcare professionals can be directly opposed to patients' wishes of active selfcare [44,45]. This may be due to limited knowledge on the part of healthcare professionals, since patients' views are well aligned with the definition of selfcare from the WHO in **Box 1.3**. Another potential explanation is clinicians' insecurity of their own role with regards to more autonomous patients, since traditionally, healthcare professionals were experts in a system organised for dealing with acute injuries and illnesses and patients were passive recipients of care.

This gap between healthcare and selfcare can become apparent to physicians who are faced with health challenges. Disease and illness are separate but related concepts, where illness is the subjective experience and disease relates to what can be observed from the outside [46]. As is demonstrated from articles by physicians who became patients [47,48], the lived experience of illness can be very different from the learned experience

of disease. One pertinent example is an article by a neurologist who developed PD himself and stated that: *"The experience has deepened and enriched my understanding of the doctor – patient relationship, and I hope it has helped me to become a better neurologist"* [49]. This apparent inability of clinicians to fully see the complete patient perspective unless they become a patient themselves, could be one explanation for the different views of selfcare between patients and clinicians mentioned above.

In the beginning of 2019, I started using a rollator when walking outside. A rollator is like a small cart with four wheels and handles and it gives me support when I get into a freezing-of-gait (FOG) episode. FOG is a strange phenomenon where my feet, for sometimes unclear reasons, suddenly stop and appear as if they are frozen to the ground.

It wasn't an easy decision to start walking with a rollator, I thought it would feel like I was giving up and letting PD win. When I started walking around town with my new rollator, I realised that I didn't feel the stigma that I expected I would. I was very surprised. To add to my surprise, the rollator has actually brought benefits that I hadn't thought of. When I started using my rollator, I found that I could walk both faster and longer than I could before, when I walked with my walking stick. I was now walking more upright and was distributing my weight more evenly over my right and left foot. I was enjoying taking walks again. I hadn't realised how much I had limited myself previously.

Technology, including online tools and services, have been identified as having the potential to contribute to health and well-being for persons with PD. In the absence of an objectively measurable biomarker for PD, one important area for technology would be to support persons with PD in registering their symptoms and other observations in support of PD selfcare. Several recent studies report that persons with PD are optimistic that technology will enable them to improve their abilities to successfully manage the progressive, variable, and unpredictable nature of PD. i.e. support their selfcare [45,50–52]. Unfortunately however, to date, technology-related development in PD does not live up to the expectations of persons with PD [53]. One potential explanation for the lack of technological support for PD selfcare can be the differing views of selfcare of patients and clinicians mentioned above. In addition, many issues remain to be solved, including privacy and security issues, and also the reliability of the developed algorithms. A major remaining challenge is to somehow couple the new insights generated by technological approaches into the existing healthcare system, such as the electronic medical records used by healthcare professionals [54].

1

Apart from the challenges of managing my pills, which in itself is a lot of work, I also have to take care of remaining physically and mentally active and ascertaining that I get enough exercise. If I don't, my muscles will tense up and ache. Earlier in my disease trajectory, it was easier to combine my other obligations and commitments with also getting a reasonable amount of exercise into my everyday life. Nowadays however, my increased symptoms means that it's more difficult to find the right type of exercise, the best support/advice, and the appropriate structure around the training that I need. For example, before I started using my rollator, I used to attend group trainings of boxing for PD but, unfortunately, these days that is much more difficult. My gait problems make group training problematic and with longer disease duration, my energy reserves are more easily depleted. This means that I have to be a bit more creative when it comes to exercise and do my best to stay as active as I can, without putting too much pressure on myself. To find the right balance, I prioritise short exercise sessions over long ones, I prefer stretching over really high-intensity training, and value safety over really challenging exercises. These are my individual choices and each person with PD must find their own exercise regime.

That our healthcare systems are inadequately organised when it comes to effectively supporting persons living with chronic conditions has been known for at least 25 years [55]. Attempts have certainly been made to address this but as the persisting gaps between healthcare and selfcare described above demonstrate, we still have a long way to go. It seems self-evident that the field of selfcare would benefit from a joint understanding of both the lived and the learned experience. Nevertheless, to date, the majority of research on selfcare has been conducted by persons without lived experience of the disease in question. Patient-led research, which I will elaborate on later in this chapter, is one way to change that.

Personal science and self-tracking

The term personal science has been used for about a decade but is a more recent concept within academia. In the way I use the term in this thesis, it was first mentioned in a scientific article in 2016 [56]. However, while the expression itself is relatively new, the phenomenon it is associated with is not. The practice of personal science, which I will describe further in the following sections, has emerged from the 10+ years of work in The Quantified Self community. The Quantified Self is a social movement that was founded in San Francisco in 2008 and now entails a global network of self-trackers that meet in local gatherings and international conferences [57,58]. The meetings focus on "first person accounts of self-tracking projects and experiments", addressing the three questions: *What Did You Do? How Did You Do It? What Did You Learn?* [57–59].

The Quantified Self community has always consisted of both academically trained researchers and lay scientists. The community is focused on knowledge production for personal use [56,60], and the presence of academically trained researchers in the community is likely to have contributed to the emergence of the term personal science.

The popular general public view of the Quantified Self community is that it is a place attracting "tech nerds" exchanging data [60]. In reality however, very little data are actually shared between people [58]. The reason for this is not, as could be imagined, data privacy concerns, but simply because people in the Quantified Self community are aware that there is more to be learned from what methods others are using than from the data of others [58]. Members of the Quantified Self community sometimes develop their own apps and other services and/or adapt apps or services, because the ones publicly available are not able to meet the users' unique individual needs [57,60]. Sharon and Zandbergen (2017) argue that the frequent critique of the Quantified Self as "data fetishism" fails to recognise the ways that the data generated by an individual can hold value and meaning to that individual that does not necessarily automatically translate to others [57]. Members of the Quantified Self community are known as self-trackers. Examples of successful projects are given in the section *Personal science projects and personal scientists* on page 30.

Personal science - a framework of study

Today's healthcare has greatly benefited from the introduction of the traditional model for biomedical research. Specifically, the randomised controlled trial (RCT) model has helped us to create an evidence base to demonstrate both the efficacy and safety of a variety of treatments for many diseases, including PD. However, we increasingly realise that the current model for evidence-based medicine has its limitations [61,62]. Examples of shortcomings of RCTs include: the populations studied in RCTs are often selected using strict inclusion/exclusion criteria and most RCTs are conducted during a limited period of time and on a relatively small group, attrition rates are often high, especially when studying non-pharmacological interventions, and implementing results from RCTs in everyday clinical practice often proves challenging [63]. This means that although the evidence generated through RCTs may be useful on a group level perspective, the behaviours and characteristics of individual patients in a real-world setting may differ significantly from the grand average of the population included in the trial.

An RCT can be said to answer the question: "Is this treatment likely to work for an average patient?" It does not tell us what will actually work for an individual patient. It is also difficult to predict whether the treatment is safe for that particular patient, and what adverse effects this individual might experience. In real life, many persons

with PD typically take multiple pharmacological as well as non-pharmacological treatments, and RCTs are rarely designed to study such complex situations. There is a need to add a complementary approach to arrive at what works for an individual patient. One possible way forward is the single subject research design. Single subject research design is a term used for all research performed with one individual participant, including case studies, single case research, and N-of-1 trials. The concept of N-of-1 trials consists of "multiple crossover trials, usually randomized and often blinded, conducted in a single patient" [64]. Several N-of-1 studies can be combined to generate group level evidence [64]. N-of-1 trials have been reported to be of variable quality [65], but when designed and conducted well, are regarded as a Type 1 level of evidence according to the Oxford Centre for Evidence Based Medicine [66,67].

Definition of personal science

Personal science can be seen as a new kind of single subject research design. Personal science has been described as: *"the practice of using empirical methods to explore personal questions"* [59] and as: *"self-directed N-of-1 studies"* [68], *"an interest in collecting data about their own bodies or lives in order to obtain insights into their everyday health or performance"* [56]. Based on these key references, in this thesis I define personal science as *"the practice of exploring personally consequential questions by conducting self-directed N-of-1 studies using a structured empirical approach."* Personal science is an overarching label for practices where the research participant is also the researcher [57–60,69]. A partly overlapping concept is citizen science, which connotes active public involvement in scientific research [70]. The main difference is that personal science is always self-directed while the majority of citizen science projects are not [59]. Important academic contributions in developing the field of personal science has been made by Roberts [71], Wolf [59,68,72,73], de Groot [59,68,74], Larsen and Blomseth Christiansen [58,75,76], Wright [77], Heyen [56,60], Li and colleagues [78], and Almalki and colleagues [79].

Personal science can be practiced at different levels of impact. At the first level, an individual uses personal science only to improve their own selfcare. The second level is achieved when the methods and/or findings are also publicly shared, for example through blogs, presentations, videos or similar approaches. Sharing can also be done via conventional academic journals. Thereby, personal science poses some new ethical challenges where some of the most pressing to discuss and resolve include: What happens when individuals move beyond actively participating in clinical research to using empirical methods to improve their own selfcare? What happens when people managing health challenges on a daily basis, also known as patients, make use of the possibilities of the Internet and other technological developments to conduct their own research? To what extent do current ethical frameworks apply to these emerging

practices? Do specific ethical challenges surface when individuals also intend to disseminate their findings by publishing their conclusions in scientific journals?

It is important to note that personal science is not intended to replace conventional clinical research, but to complement and enrich its practices, results, and applicability. Though in some ways personal science may appear comparable to clinical N-of-1 trials, the two are distinctly different. The aim of clinical N-of-1 trials is to combine providing personal benefit to individual patients while being clinically practical as well as generating knowledge that can be publicly disseminated and can be applicable beyond the patient in the trial. In personal science, the aim is to find meaningful answers that matter first and foremost to an individual with a particular health challenge. The most important difference, however, lies at the level of supervision and control: for clinical N-of-1 trials, research is conducted by a researcher on an individual patient. In contrast, for personal science, the researcher and the participant are one and the same. Personal science has limitations as well as potential, for example, personal science is not suitable for conducting research aiming to primarily determine effects on a group level.

I have been a self-tracker since before I knew there was a word for it and I also write about it on my personal blog (https://www.riggare.se). I use self-tracking tools to better understand the variations of my PD but also for other aspects of my life. My engineering training has provided me with the mind-set suited for observing phenomena, evaluating, and adjusting, repeating as necessary.

When I attended my first Quantified Self conference in 2011, I immediately felt at home in the community. For a long time, I didn't realise what made it so special but when comparing it to some of the other conferences I have attended, it was clear: at Quantified Self conferences I forget that I have PD!

People there are not interested in what disease I have; they're interested in what I do to deal with my situation. What kind of data am I capturing? What observations am I making? What tools do I use? What are my hypotheses? I am seen as an autonomous person with the ability to reason and learn. That is a very stark contrast to some of my healthcare encounters, where I feel like I am seen as completely incompetent, simply because I happen to be a patient.

I also attend patient-centred conferences focused on PD, and I learn a lot about PD, available treatments and ongoing clinical research. It can be a relief to be among people who deeply understand what PD feels like. But we all also compare our own parkinsonian symptoms or signs with those of others around

us: "Is that person's PD more severe than mine?" "Does that person have this or that symptom?" This comparison can be helpful to understand my own condition and situation, but it can also emphasise the disease.

Sometimes it's just nice to be able to forget I have PD.

Self-tracking

There is no generally agreed upon and unambiguous definition of **self-tracking** [80]. In this thesis, I define self-tracking as a method for data collection and *"a process of deliberately collecting and structuring observations about one's own life"* [59]. I use the term to signify the most frequently used method for data collection in personal science. The method can also be used for collecting data in conventional clinical research on groups of participants.

It is generally agreed that self-tracking is an important element of selfcare in many chronic conditions; examples of self-tracking can involve persons with diabetes measuring their blood glucose levels or persons with asthma monitoring their peak respiratory flow [44,81]. Self-tracking can be aided by digital technologies but can also be done simply using pen and paper. In 2013 Pew Internet reported [82], that self-tracking is very common. Specifically, 69% of U.S. adults track one or several measures connected to health, and people with chronic conditions are more likely to track in an organized way. Among the people with chronic conditions in the study, 45% used paper and 22% used some sort of device [82].

Rapid technical developments have led to the introduction of advanced medical technologies for self-tracking that were previously only available to trained medical professionals, e.g. ECG devices, increasingly available to consumers. For example, widely available tools such as smartphones with their embedded sensors and specific health apps, as well as smartwatches and other types of body-worn sensors, can now be used by individuals to facilitate data collection about their lives and their own health [83,84]. A recent study found that the use of wearable technology among healthcare consumers is growing: from 9% in 2014 to 33% in 2018 [85].

However, I want to emphasise that self-tracking using technology also comes with a number of important limitations. For example, the accuracy of commercially available algorithms has often not been evaluated under carefully controlled experimental conditions. Furthermore, to protect the proprietary rights, background information on the algorithms of devices (let alone the algorithms themselves) is rarely shared publicly. Serious errors sometimes come to light. For example, Fitbit's heart rate algorithm proved

to have a poor correlation with gold standard values, provided by heart rates recorded using an ECG [86]. Patients also face privacy and security risks with data collected using commercially available devices. Individuals may not always realise that the data, which they are collecting using commercial technologies to promote their own health, may also be utilised by other parties, who may not always have the user's best interest in mind. And finally, as I will also address in this thesis, there may be a limit to what can be expected from people living with a chronic condition. Sometimes medical professionals think that they can readily defer the tracking of symptoms or treatment effects to participants in the studies, thereby relieving the pressure on the medical system. While this may be understandable from their perspective, it is all but straightforward to think that each and every patient is able and willing to regularly engage in self-tracking. One reason is that it may confront them with their disease on a much more continuous basis. I will address this and other concerns and limitations of self-tracking in this thesis. Definitions of personal science and self-tracking are summarised in **Box 1.4**.

BOX 1.4: Personal science.

Personal science and self-tracking
Personal science is a framework of study for single subject research defined in this thesis as: *"the practice of exploring personally consequential questions by conducting self-directed N-of-1 studies using a structured empirical approach."*
Self-tracking is a method for data collection defined as *"a process of deliberately collecting and structuring observations about one's own life"* [59]. It can be used in personal science as well as in conventional clinical group research.

Applications of self-tracking

Research from the field of oncology indicates that self-tracking can significantly improve clinical outcomes; in an RCT of 766 patients undergoing chemotherapy, overall survival for patients tracking 12 symptoms using a web-based platform was compared to care as usual [87]. Patients who tracked their symptoms had a median overall survival of 31.2 months compared with 26.0 months for the group receiving only usual care. The main reasons stated for this difference were early responsiveness by nurses with respect to possible adverse events, and increased tolerability of the chemotherapy for the patients through the process of tracking. According to an online commentary by Eric Topol [88], this median survival improvement is of the same magnitude as what can be achieved with for example immunotherapy, which would cost at least $100,000 per treatment. I should point out that other explanations also are possible: perhaps people who actively comply with a web-based monitoring system are also the ones who generally lead a healthier lifestyle, and also comply better with e.g. lifestyle interventions such as exercise or a healthy diet. But the results are promising and deserve further study.

Previous research demonstrates that self-tracking can be valuable also to persons with PD. A study of 75 persons with PD in Australia showed that the frequency of non-technology based symptom self-tracking (observing five pre-decided symptoms) was significantly correlated with better selfcare [27]. In a Swedish study of the effects of selfcare education, 95 persons with PD and 55 care partners were randomised to receive normal care or to participate in a selfcare education program along with normal care [89]. The selfcare program focused on non-technology based self-tracking as a tool for strengthening the ability to live a good life with PD. Evaluation of the program showed significant improvements among persons with PD who participated in the selfcare education in health status, skills, attitudes, and learning compared to the group that received only normal care [89].

There is also research support for the proposition that self-tracking using technology can be beneficial for persons with PD. In a small study, 22 persons with PD were randomised to either wear sensors for tracking movement (registering tremor, dyskinesia/hypokinesia, and gait) or not to wear sensors [90]. In the last 8 weeks of the 12-week study, the sensor group was given feedback based on their individual sensor data, and that group improved in mobility compared to the non-sensor group. The difference was reported to approach significance at 12 weeks and was found to be significant at follow-up [90].

These examples (one in cancer and three from the field of PD) suggest that individuals can benefit from learning about their own situation by using self-tracking (technology based or non-technology based). The process of creating feedback loops to self-tracking individuals based on their own data can facilitate learning, improve selfcare and have beneficial effects on their health.

Personal science projects and personal scientists

Given that personal science is an emerging research field, it is only natural that the current evidence base is small. It is however increasing rapidly, both in volume and scientific rigour. For example, in 2017, a focus theme of the scientific journal *Methods of Information in Medicine* enabled personal scientists to publish their work as "personal science reports" [68]. The foundation of the field consists of the joint practices of the global Quantified Self community. This practice has led to more than 1,000 recorded presentations on www.quantifiedself.com, many of which have been transcribed [58,59]. This archive has formed the basis for several ethnographical/sociological studies of the Quantified Self community [57,91–93].

Examples of personal scientists who have succeeded in making discoveries of personal importance to themselves include Larry Smarr, who was able to self-diagnose his

inflammatory bowel disease from gut microbiome analyses [94] despite his physician's resistance [57] and Dana Lewis, who aims to help both herself and a wider community of persons with type 1 diabetes by developing tools and methods to achieve improved blood glucose control [95]. I want to emphasise that success in personal science does not necessarily mean that the individual insights are also applicable to a larger group. Other well-known personal scientists are Thomas Blomseth Christiansen, who has used personal science methods to manage severe allergies and eczema [58], and Anne Wright, who systematically explored correlations between food intake and symptoms [77]. Both of them have an engineering background: Blomseth Christiansen as a software developer, and Wright was the NASA lead systems engineer for the Mars rover project. They have developed personal science practices to successfully manage their health challenges and both describe their exploratory process using the engineering term debugging [58,77]. What all these examples have in common is that their main contributions have been on developing methods for personal science.

Along with these examples of single-subject studies there have also been efforts to develop group-based personal science projects. In a study of 20 personal scientists, every person conducted their own single subject study, exploring individual questions around blood cholesterol and triglycerides [72]. Additionally, they all discussed their individual findings on a group level [72]. The study found that individuals using self-collected lipid data can contribute to their own health as well as to generalisable health knowledge [72].

It has been suggested that persons using personal science in collaboration with clinicians are in a better position to achieve a sustained behavioural change [96] and it has been shown to be important to focus on the person's individual goals [97]. There is a need for more research into the field of personal science [59,97], in order to better understand both the possibilities and opportunities, and the risks and challenges.

Patient-led research

An increasing body of evidence demonstrates that the priorities of patients, across diseases, are often different than what clinicians think they are [98,99], also in the field of PD [23,100,101]. This means that when clinicians and researchers set priorities for research based on *their perception* of what patients think is important, they are often not addressing the issues that really matter most to patients. Estimates suggest that 85% of biomedical research results in avoidable waste, one of the main reasons being a lack of relevance to patients [99]. Patients engaging more directly in the research process can improve research in terms of quality and relevance to patients [102–104]. Involving patients more closely in defining the research agenda, in designing research studies and in helping to interpret the findings is a process that has been referred to as

participatory science [105]. But this is still not the same as truly patient-led research.

In this thesis, I define **patient-led research** as research led by **patient researchers**; persons with lived experience of a disease, disability, or another health challenge who are openly using those experiences in doing research, within academia or in other contexts. This can also include a family member or partner of the person living with a disease. Patient-led research is a way to bringing the lived experience of illness into the learned experience of diseases, thereby potentially improving the relevance of conducted research. The field of patient-led research is not yet unambiguously defined; other terms used include for example participant-led research [73,106], parent-led research [107], or patient-driven research [108]. These terms are used to describe work done with different degrees of involvement of patients and/or persons close to them.

In 2010 I decided that I wanted to combine my engineering training with my patient experiences and try to improve things for myself and others with chronic diseases. I wanted to help patients and researchers understand each other better. I had realised that in order to be taken seriously by academic researchers, you had to be a researcher yourself. And I was sad that I wasn't. My husband said that maybe it wasn't too late to go into research and start working on a PhD. At first. I rejected his suggestion as unrealistic, but the idea grew on me. I set out to explore my own experiences of living with a complex disease in a PhD project, and eventually, this led to my starting to refer to myself as a patient researcher.

The ongoing COVID-19 pandemic has demonstrated what can happen when a completely new disease emerges in the age of the many possibilities offered by the internet and by the many technological developments that are beginning to emerge in the medical field. The exact numbers are still unclear but it has been reported that about 10% of patients who are infected will develop "long-term COVID" (experiencing symptoms of a COVID-19 infection for longer than 3-4 weeks) [109]. Thanks to social media and the internet, people living with long-term COVID started connecting with each other and a self-organised online support group emerged [110]. The need for more knowledge about this new and unknown condition led members of the patient community with previous experience from research work to design an online survey. The study was completed in a very short time: it was initiated, designed, data were collected, analysed, and reported in less than six weeks [111]. The article reporting the study was self-published on a website (https://patientresearchcovid19.com) and not in a peer-reviewed journal. Nevertheless, the study has been cited several times in articles in high-impact scientific journals, one of which being a "practice pointer"

in the BMJ, giving recommendations on the management of long-term COVID in primary care [112]. The long-term COVID patient researchers' work has also been used to inform a more conventional academic study on long-term COVID [109]. This is an excellent illustration of how the lived experience of a condition can directly contribute to scientific development. At the same time, this example also underscores the need for patient communities to collaborate intensely with clinicians and researchers. There is considerable debate as to what the exact nature is of the long-term COVID syndrome, and while patients are better positioned than anyone else to describe the nature and the impact of the syndrome, it will likely require careful research and the eyes of experienced clinicians to identify the various underlying sources of disability in persons experiencing the long-term COVID syndrome.

Another example of research led by patient researchers includes personal scientists like Dana Lewis and the #WeAreNotWaiting movement in type 1 diabetes [113]. Also, researchers using their lived experience in a traditional academic environment like Rosamund Snow, who used her own experiences of managing type 1 diabetes in conducting her PhD studies involving persons with diabetes [44]. Patient researchers in academia often receive strong pushback, for example when Rosamund Snow went from being Ms Snow to Dr Snow, she was no longer allowed to be a patient representative in groups working in patient and public involvement in research on the grounds of her no longer being a "real" patient [114]. She offers three potential explanations: real patients 1) are not supposed to have knowledge, 2) do not have the ability to acquire knowledge, and 3) need to be protected from academics.

For patient researchers working within academia, it is beneficial (or almost necessary) to be academically trained, and this can take place prior to becoming a patient or after.

I started my PhD at Karolinska Institutet in March of 2012 and it's been all but straight-forward. To be honest, if I had known then what I know now about all the issues I would encounter, I'm not sure I would have gone through with it. Of course, the issues that most PhD students inevitably face were not surprising, like having your submitted manuscript – after all the hard work of writing up your study findings – receiving a 'desk reject' from a journal editor, or the complex process of struggling with wrapping your head around the complicated rules for required academic credits.

What was more unexpected was the resistance from the academic system itself. An example is when I was discussing my research with a senior academic researcher and he said: "But Sara, why would patients want to do research on

themselves anyway? Wouldn't it be better if you all just give your data to a proper researcher?"

I didn't know how to respond. What did he mean "proper researcher"? It was 2018 and I was hoping to be able to defend my PhD thesis soon. His comment shook me. In a way, I can understand where he was coming from: in conventional research, patients are just participants in research studies, they are not researchers. "But", I thought, "have we really not come farther than this by now?"

I strongly believe that the best way to change things is to actively start doing things differently.

There are also examples of active patient researchers within academia in the field of PD. These active patients embrace the full spectrum of the lived experience of PD and also openly share their perspectives, thus contributing to influencing research [23,115–120].

OUTLINE OF THIS THESIS

This thesis contributes to the understanding and development of selfcare in PD by examining how patient-led research in the form of personal science can contribute to selfcare in PD.

In **Part I**, I will explore the feasibility of using personal science for selfcare in PD by presenting two single subject studies where I use myself as the research participant. In **Chapter 2**, I will present a study with an observational design. In **Chapter 3**, I will use an interventional design. In both chapters, I will elaborate on how personal science can contribute to PD selfcare.

In **Part II**, I will investigate the transferability of the methods of personal science to more general users, represented by my fellow persons with PD in Sweden. In **Chapter 4**, I will explore the current state for selfcare for persons with PD in Sweden. In **Chapter 5**, I will investigate the experiences and opinions of persons with PD in Sweden on personal science.

In **Part III**, I will examine some aspects of patient-led research. In **Chapter 6**, I will briefly describe the different roles patients can take in research and discuss some of the opportunities and challenges associated with patient researchers. In **Chapter 7**, I will compare the ethical aspects of using self-tracking for personal science in PD to using self-tracking in the context of conducting clinical research on groups of research participants.

In **Part IV**, the findings are summarised (**Chapter 8**) and discussed, including future perspectives (**Chapter 9**).

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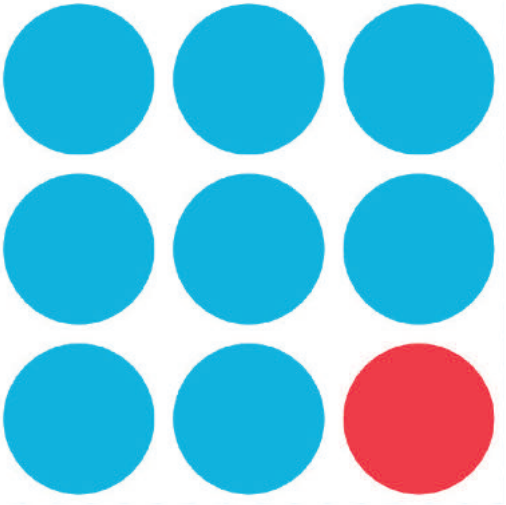
PART I

EXPLORING FEASIBILITY OF PERSONAL SCIENCE IN PARKINSON'S DISEASE



Since I am absolutely dependent on my pills for my body to function, I have to pay attention to always having sufficient amounts of the correct combinations with me. Every few weeks I prepare my pills for the upcoming weeks. This means that I take out a large tray that I put my different pill organisers on and distribute the right combination of pills into each compartment. I often start with my morning doses, which comprise three different drugs. For the other doses I need containers that are not too big (I want them to easily fit into my handbag) and also not too small (I don't want to have to refill them every day). They also need to be easy to open and close but not too easy (I don't want all the pills to fall out in my handbag). I use a transparent plastic pill organiser consisting of five separate cylinders that can be screwed together to form a longer cylinder where one cylinder acts as the lid to the previous one. Each of the five containers can hold two days' worth of rest-of-the-day doses which means that I can easily have up to ten days' worth of pills (not counting morning doses) in my handbag. But why don't I simply bring my pills in their original packaging? Surely that must be easier? No, it's actually not, for a number of reasons: 1) It can be surprisingly difficult to remember if you've taken a dose or not when you're busy working, at an important meeting, or simply out and about. If I preload each cylinder with two days' worth of rest-of-the-day doses, I can easily check how much is left if I am uncertain. 2) Original packaging often holds 100 pills and if I were to have several of those in my handbag all the time, I would need to have a very big handbag. 3) When it is time for the next dose, my fingers have started getting stiff and fumbly and if I didn't have the pills reasonably well organised in the combinations I am meant to take, I would risk dropping a pill or two.

NOTE

As is pointed out in **Chapter 1**, personal science is an emerging research field that can be seen as a new kind of single subject research design. The fact that the field is new means that when the study reported in this chapter was conducted, slightly different terminology was used.

In this chapter, the term *patient-initiated self-tracking* should be read to mean *personal science*. When the term *self-tracking* is used alone, it should be read to mean a method for data collection.

CHAPTER 2

Personal science in PD: observational design

Published as: Precision Medicine in Parkinson's Disease - Exploring Patient-Initiated Self-Tracking. Riggare S, Hägglund M. J Parkinsons Dis. 2018;8(3):441-446.

ABSTRACT

Background

Individually tailored healthcare, in the form of precision medicine, holds substantial potential for the future of medicine, especially for a complex disorder like Parkinson's disease (PD). Patient self-tracking is an under-researched area in PD.

Objective

This study aimed to explore patient-initiated self-tracking in PD and discuss it in the context of precision medicine.

Methods

The first author used a smartphone app to capture finger-tapping data and also noted times for medication intakes.

Results

Data were collected during four subsequent days. Only data from the first two days were complete enough to analyze, leading to the realization that the collection of data over a period of time can pose a significant burden to patients. From the first two days of data, a dip in finger function was observed around the time for the second medication dose of the day.

Conclusions

Patient-initiated self-tracking enabled the first author to glean important insights about how her PD symptoms varied over the course of the day. Symptom tracking holds great potential in precision medicine and can, if shared in a clinical encounter, contribute to the learning of both patient and clinician. More work is needed to develop this field and extra focus needs to be given to balancing the burden of tracking for the patient against any expected benefit.

INTRODUCTION

Precision medicine (or personalized medicine) is achieved when "health care is individually tailored on the basis of a person's genes, lifestyle and environment" [1]. A complex and multifaceted neurodegenerative disorder like Parkinson's disease (PD), with its plethora of symptoms (motor and non-motor) and treatment side effects, is likely to benefit from a precision medicine approach. Such an approach would include consideration of for example genes, clinical subtypes, personality and preferences, lifestyle, pharmaco-economics, aging, and comorbidities [2,3]. Technology-based objective measures are seen as a potential contributor to precision medicine in PD [4]. To achieve precision medicine, focus needs to shift from patient cohorts to what works for an individual patient [5,6]. Historically, important insights have come from observations made by clinicians on individual patients or on themselves. The former is often reported as a case study and the latter is known as self-experimentation. Before the Helsinki Declaration, physicians conducting experiments on themselves prior to testing their ideas on patients was one way to ensure research ethics [7]. Self-experimentation has resulted in important progress including a number of Nobel prizes [8], for example for the discovery of *Helicobacter pylori* as a cause for gastric ulcers [9]. Observations by patients have also contributed to important insights. A few notable examples are the surprising side effects of Sildenafil or that smoking cessation is associated with depression, both of which has led to new effective treatments [10]. Case reports are frequently used in PD to report on clinical observations or findings and we have previously published a patient-driven study, in which the first author (SR) conducted an experiment on herself to improve self-management of her PD [11][3]. To the best of our knowledge, no other patient-driven case reports have been published in PD.

Technology enables patients to be more active in the management of their health by making use of technology for self-tracking of symptoms and selfcare [12]. Self-tracking can also significantly improve clinical measures; in an RCT of 766 patients undergoing chemotherapy, overall survival for patients tracking 12 symptoms using a web-based platform was compared to care as usual. Patients tracking symptoms had a median overall survival of 31.2 months compared with 26.0 months for the group receiving usual care. The main reasons for the difference are early responsiveness by nurses on potentially adverse events, and increased tolerability to chemotherapy for the patients through the process of tracking [13].

Patient-initiated self-tracking is an under-researched area in PD. A review of sensors for patient self-tracking found that technology-based objective measures have been

3 Chapter 3 of this thesis

demonstrated to have clinical relevance for assessing motor aspects of PD as well as the potential to improve individual outcomes [14]. Efforts have also been made into discussing the potential of using technology to quantify well-being and HRQoL in PD [15]. The latter is a viewpoint article and also includes the patient perspective in the form of three persons with PD sharing what they hope technology can contribute to their PD management. We have however not been able to find any studies on patients' practical experiences of applying technological self-tracking to aspects of PD.

The primary objective of this study was to explore patient-initiated self-tracking in PD by investigating the daily variations of the effects of the first author's PD medication. The secondary objective was to discuss this example in the context of precision medicine.

METHODS

SR displayed her first symptoms of PD in her early teens, around 1984, and was formally diagnosed with juvenile onset PD in 2003. Her engineering background and interest in technology has led to exploration of self-tracking for personal benefit. Her most affected side is the left and main symptoms are bradykinesia and rigidity. She does not have tremor and was on a stable dose of PD medication during the study period (see **Table 2.1**).

Tapping tests are frequently used in neurological clinical examinations to assess different aspects of motor function. They have been shown to be a useful, reliable and valid tool for clinicians to assess symptoms of advanced PD [16]. At home touch screen assessments of finger tapping have been shown to correlate to conventional scales for evaluating PD [17]. Our study was inspired by a Swedish study [18] using a personal digital assistant (PDA) for capturing finger tapping during 20 seconds of alternate tapping, where the user taps with the same finger but alternates between two boxes, side by side, to tap in. In that study, patients were expected to perform the test in a home environment several times per day for a period of about one week. The system has demonstrated the ability to capture individual daily variability of PD [19].
At the time for data collection (March 2012), the subject/first author (SR) owned an iPhone 4 to use for capturing finger tapping data. Therefore a search was made of the Apple app store for a free iOS app that was able to capture alternating finger tapping for at least 20 seconds. The app 'FastFingers' was found to record the number of taps on the screen for 30 seconds from the first tap and present the total number on the screen. We could not find any apps designed for alternating tapping so we decided to use FastFingers with non-alternating tapping.

TABLE 2.1: Medication regimen for the study period (names given as name of active substance(-s) with Swedish brand names in brackets).

Medication timings	6AM	11:30AM	3PM	6:30PM	9PM	10:30PM
Levodopa/Carbidopa, 100/25 mg (Madopark)	¼	¼	¼	¼		¼
Ropinerol controlled release, 2 mg (Requip depot)	2		1		1	
Entacapone, 200 mg (Comtess)	1		1	1		
Rasagiline, 1 mg (Azilect)	1					
Levothyroxine, 100 microg* (Levaxin)	1					

* Levothyroxine is taken as a consequence of a thyroidectomy in 2000.

The intention was to conduct tapping tests frequently, at least 10 times per day with each hand during a study period of 5 days. During the study period, times for medication intakes were noted manually and finger tapping was captured using the iPhone app FastFingers. Data was collated and visualized using Microsoft Excel.

Ethical considerations are important in all research and although an ethical review board has not reviewed our study, ethical issues have been considered. The subject, who is also the first author, conceived the study. She was well aware of the potential risks and benefits and the usual power imbalance between researcher and research subject does not apply. This experiment was originally conducted without the intent to publish the results. However, we consider it ethical to share our experiences and lessons learned in order to further the field of patient-initiated self-tracking by publishing our findings in a scientific journal.

RESULTS

Data collection was initiated on 12 March 2012 and continued for four days. Finger tapping was conducted 13 times with each hand on the first day, 9 times on the second, 7 times on the third, and 3 times on the fourth. On the fifth day no data was collected. SR realized that to add the collection of data to your everyday life, even if it is self-imposed, takes time and effort and can be a challenge to combine with work, family life and other obligations.

Only data from the first two days were complete enough to analyze. Data from finger tapping tests performed on 12 March and 13 March are presented in **Figure 2.1**. The graph was produced in Microsoft Excel and shows tapping test results (lines) and medication intakes (bars).

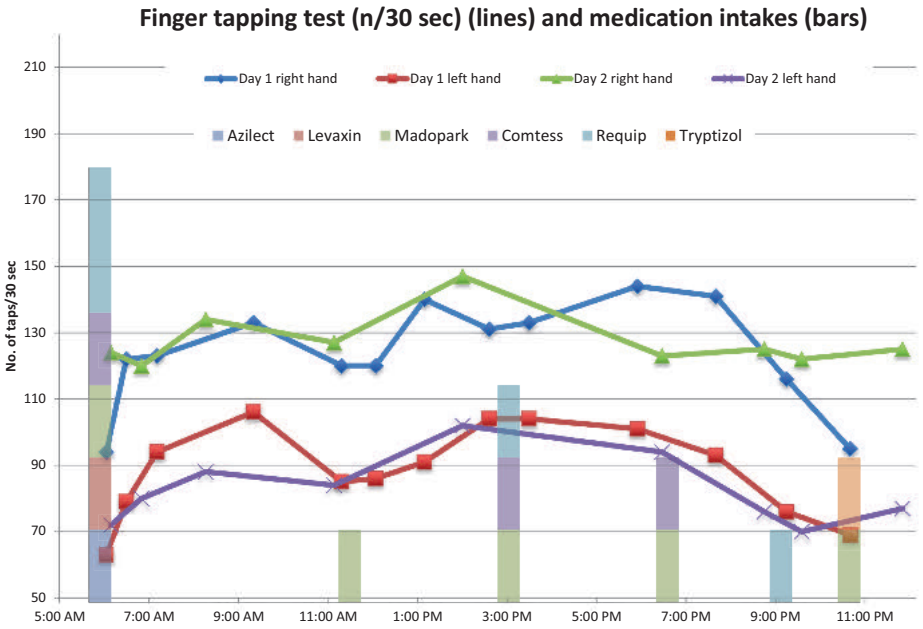


FIGURE 2.1. Lines show finger tapping results (right and left hand) from 12th (day 1) and 13th (day 2) of March 2012. Bars show medication intake times.

Reflecting on the results of the two days led to the observation that there seemed to be a dip in finger function, especially on the more affected, left side, around the time for the second dose of the day (11:30 am), see **Figure 2.1**. The results are 85 taps with the left hand in 30 seconds at 11:19AM on the first day, to be compared to the maximum value that day, which was 106 taps at 9:21AM.

Based on the results of the test, SR initiated a dialogue with her neurologist about adjusting the timing and/or dosage of medications to reduce the dip seen in **Figure 2.1**.

DISCUSSION

The future of medicine lies in precision medicine and its potential for contributing to personalized treatments. The individualized manifestations and fluctuating nature of PD should make it particularly suitable for a precision medicine approach, including patients' self-tracking efforts with the aim of enabling better understanding of individual variations.

The subject of our study realized with the help of a finger tapping test that her motor function was not the same throughout the day and specifically that she had a dip in function around lunchtime. The question arises if such a dip is clinically relevant. The study was patient-initiated, and no clinicians were involved in collecting, analyzing, or interpreting the results. We do however consider it reasonable to assume that a difference in finger agility of 21 taps per 30 seconds is likely to have an effect on for example the subject's typing speed, which is of relevance to the subject in question.

Another relevant question would be: Is two days of data enough to find patterns in disease and medication effect? Would not a longer period of data collection be able to provide more valuable insights for both patient and physician? Of course, more data is always desirable and we cannot be certain that the patterns identified would be seen also on the subsequent days. From a patient perspective, identifying a problem or seeing an effect may often be considered enough, and weighing the added benefit of gathering more data against the burden of continuing the data collection may result in patients terminating data collection earlier than what would be the case in a conventional clinical study. The primary objective of this study was to explore patient-initiated self-tracking in PD and all findings will need further work to confirm or dismiss their importance.

There also seems to be other potential benefits to self-tracking. SR found, similar to what Pantzar and Ruckenstein [20] reported from their participants, that when she interacted with the collected data during analysis and visualization, she was able to reflect on what they meant in her specific context. The feedback loops that were created led to SR learning about both the specifics of her different medications as well as what the combination of them meant for her motor function over the course of the day. When data from self-tracking is shared in a clinical encounter, it has the double benefit of potentially contributing to the learning of both patient and clinician.

Since March 2012, when the data for this study were collected, there has been a lot of development in the area of smartphone aided self-tracking for PD. A few notable examples are the smartphone apps mPower and uMotif, and the SENSE-PARK system. The mPower app was launched as part of Apple Research Kit in March 2015, aiming towards a better understanding of the variations of PD and potential modulators as well as providing real-time feedback to the participants [21]. Publications from research using mPower show interesting results and great potential but as far as we can understand, real-time feedback has not yet been achieved [22–24]. The self-management app uMotif (available on iPhone and Android) was evaluated in a 16 week trial of 158 persons with PD where using the app was compared to treatment as usual. A small but statistically significant improvement was seen in self-reported medication adherence and persons

with PD perceived increased quality of consultation (other outcome measures were QoL and symptom control). The SENSE-PARK system was developed in an EU project for home-based evaluation of PD and consists of software, a smartphone app, a Wii balance board and a set of sensors. The feasibility and usability of the system tested positively in a 12 week study of 22 persons with PD [25]. The funding period of the project has ended, and we have not been able to ascertain the current status of the system. Evaluations of the patient perspective seem to not have been the primary focus in these systems. The only example we have been able to find exploring the user perspective is a formative evaluation of the consent process of mPower [26], which is of little relevance to our study, since consent is given a limited number of times.

Limitations

Our study is not without limitations. The test we used does not give any information regarding the non-motor symptoms of PD. In the case of our user, SR, this may not be a major issue, since she does not experience a lot of non-motor symptoms. Complementary tests and/or devices can be added if other symptoms need to be investigated. Furthermore, we have only explored self-tracking for persons with PD in the context of medication effect. Other potential uses, for example tracking the effects of exercise or diet could be investigated in future studies.

There are also advantages and limitations associated with our chosen study design. A case study is able to describe an observed effect or phenomenon in one or a small number of subjects in a timely manner in order to offer the scientific community the opportunity to discuss and critique the findings. The results from SR's self-tracking can however not be extrapolated beyond the subject in this study without further research. One can also question whether SR is representative for the larger population of people with PD. Not everyone will have the skills, knowledge and attitude to do this type of self-tracking, however we hypothesize that with improved tools and awareness of the benefits of self-tracking an increasing number of people with PD would be able to apply such approaches. Another important limitation for broader application of approaches of this kind is that they require patients with a certain level of autonomy that have the necessary abilities and skills to handle technology.

CONCLUSIONS

In conclusion, this study shows that a smartphone finger-tapping test enabled the subject in the study, also the first author, to glean important insights about how the effects of her PD medications varied over the course of the day. This suggests that symptom self-tracking may be useful to persons with PD. These variations in medication

effect could not have been discovered easily in another way and observations like these can be used as a basis for medication adjustments in collaboration with a physician. If shared in a clinical encounter, it has the double benefit of potentially contributing to the learning of both patient and clinician. The patient perspective can also be valuable when developing and implementing new healthcare technologies.

Symptom tracking holds great promise in the field of precision medicine and more work is needed to develop this field. If done right, technology has the potential to improve the understanding of PD from the perspective of all stakeholders. A major challenge when it comes to addressing the needs and wishes of all stakeholders is that the views of clinicians and researchers regarding what is important in order to help persons with PD are often different from what persons with PD consider most important [27–29]. This means that extra focus has to be given, especially in the area of self-tracking, to ensuring that new technological applications are developed to address the issues that patients find important rather than the ones clinicians prioritize. Furthermore, the burden of tracking for the patient needs to be considered in relation to the potential benefit. This will be a key factor in ensuring long-term adoption and use of self-tracking as a sustainable part of precision medicine.

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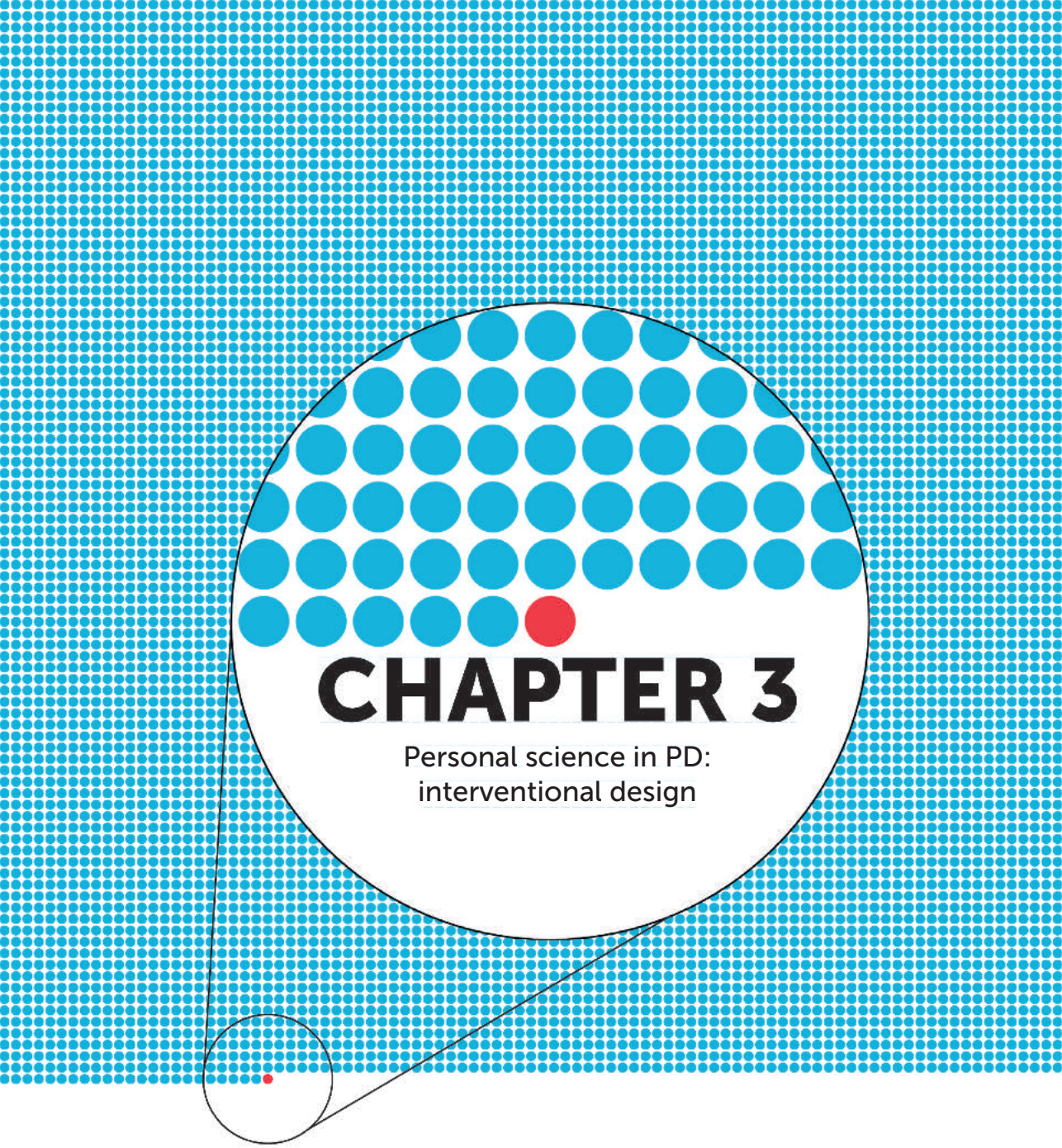
NOTE

As is pointed out in **Chapter 1**, personal science is an emerging research field that can be seen as a new kind of single subject research design. The fact that the field is new means that when the study reported in this chapter was conducted, slightly different terminology was used.

In this chapter, the term *patient-driven N-of-1 study* should be read to mean *personal science*. When the term *self-tracking* is used alone, it should be read to mean a method for data collection.



CHAPTER 3

Personal science in PD: interventional design

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Methods Inf Med. 2017 Oct 24;56(99):e123-e128.

ABSTRACT

Background

New insights and knowledge in biomedical science often come from observation and experimentation. Methods traditionally used include self-experimentation, case reports, randomised controlled trials, and N-of-1 studies. Technological advances have led to an increasing number of individuals and patients engaging in self-tracking. We use the term patient-driven N-of-1 for self-tracking performed with the explicit intention to disseminate the results by academic publishing.

Objectives

The aim of the study was to: 1) explore the potential role for patient-driven N-of-1 studies as a tool for improving self-management in Parkinson's disease (PD) using the example of managing levodopa-induced dyskinesia (LID) with nicotine, and 2) based on this example; identify some specific challenges of patient-driven N-of-1 studies.

Methods

We used a placebo-controlled patient-driven N-of-1 study with nicotine administered via e-cigarette to treat LID. The first author initiated and conducted the experiment on herself and noted her observations. The evaluations of the potential of N-of-1 for improving self-management of PD as well as the effects of nicotine on dyskinesia were based on the perception of the subject. During the planning and undertaking of the experiment, notes were made to identify challenges specific to patient-driven N-of-1 studies.

Results

The subject was able to distinguish a decrease of her LID from nicotine but no effect from placebo. The main challenges of patient-driven N-of-1 studies were identified to be associated with planning of the study, recruiting a suitable research team, making sure the data collection is optimal, analysis of data, and publication of results.

Conclusions

Our study indicates that nicotine administered via e-cigarette may have an effect on levodopa-induced dyskinesia in individual patients with PD. The main contribution is however highlighting the work done by patients on a daily basis for understanding their conditions and conducting self-tracking experiments. More work is needed to further develop methods around patient-driven N-of-1 studies for PD.

INTRODUCTION

New insights and knowledge in biomedical science are gleaned from observation, hypothesis generation and careful experimental testing. While large-scale empirical studies garner the most attention in research, the methods and knowledge that enabled large-scale studies to be conducted in the first place are often overlooked. Historically seminal ideas have occurred from a single person's careful observation and even self-experimentation. Sometimes, prior to testing an intervention on intended populations or healthy volunteers, clinicians and researchers do self-experimentation, considering it an important element of ethical research [1]. Self-experimentation (or auto-experimentation) has led to important discoveries and also a number of Nobel Prizes [2].

Experiences from single cases in the form of case reports have been used for a long time in medical education and training. From a historical perspective, descriptions of clinical cases have been found in ancient Egyptian medicine, as well as in the practices of Hippocrates and Galen [3]. Today, case reports are valued for hypothesis generation and for identifying outliers that do not fit existing models, theories, or known mechanisms. Case reports are used retrospectively to report on discoveries relating to for example new diseases, unusual manifestations of diseases or unexpected treatment side effects [4]. While case study findings are generally considered less broadly applicable across populations, researchers are beginning to acknowledge case studies as an important complement to traditional large scale empirical studies [4,5]. Large scale randomised controlled trials (RCTs) are still considered the gold standard of clinical research. There is however an increasing awareness of the limitations of RCTs, especially when it comes to applying the results to individual patients. One only has to note the sometimes vast differences in efficacy and/or side effects between the published results of new pharmaceutical trials and actual clinical practice. One possible solution may be using N-of-1 studies [6,7]. In N-of-1 studies, also referred to as single subject studies, the investigation is focused on the results from one individual, often aiming to see whether a specific intervention is effective for this individual [6,8,9]. N-of-1 studies may be especially suitable for diseases or conditions with large individual variability.

One such condition is Parkinson's Disease (PD), which is the second most common neurodegenerative disease. It causes a large variety of motor and non-motor symptoms, resulting in significant burdens on the individuals with the condition, their family members and society [10]. The condition is highly individual in nature and requires that each person with Parkinson's disease collaborate closely with their clinicians to find their optimal regime, often consisting of multiple pharmacological and other treatments [11,12]. Typically, age of onset is about 60 years but 3–5% of

persons with PD experience onset before the age of 40 [13,14], often referred to as Young Onset Parkinson's Disease (YOPD). The number of persons with PD over the age of 50 in the world exceeds 5 million (2005) and the number is predicted to double between the years 2005 and 2030 [15].

Long term use of anti-parkinsonian drugs can lead to development of a potentially debilitating and socially disruptive side effect called levodopa induced dyskinesia (LID) [16]. LID manifests as abnormal involuntary movements with writhing and sometimes jerky movement pattern. The risk of LID is significantly higher for YOPD [12–14] and can also be difficult to manage. There are only a few approved available therapeutic options with limited patient success [12].

While clinical research is traditionally initiated by clinicians or researchers, new modes of research are unfolding. Emerging technologies enable an unprecedented data collection, which has been utilized in conventional research as a tool for data collection e.g. Apple ResearchKit [17,18]. Furthermore, the prospect of learning from data collected by patients, e.g. persons with PD, in their daily lives using for example wearable devices and/or smartphones for improving clinical management and research is attracting growing interest [19–25]. Technology, such as for example smartphones, apps, sensors and other devices can also be used to facilitate data collection for individuals wanting to find answers to their own questions. This form of study can be called self-tracking [26–29], self-experimentation [30,31], Quantified Self [32] or self-quantification [33,34] and may also be done without technical assistance. A U.S. study reports that 69% of American adults track an indicator relating to health for themselves or a family member [35]. When exploring the mode of tracking, the same study shows that the use of technology is not mandatory for tracking: 49%, track 'in their heads', 34% use pen and paper, and 21% track their health using some form of technology.

Self-tracking can be applied to any personal aspect of life, but in the context of this article we focus on health or illness related factors. It can be seen as a parallel to the self-experimentation, N-of-1 studies and case reports utilized by medical professionals to learn from data or information emanating from one or a few individuals. In this article, we consider self-tracking to include any form of data-collection, observation or experiment made by an individual with or without the use of technology, concerning aspects relating to their own health or disease. We will use the term *patient-driven N-of-1* for self-tracking performed with the explicit intention to disseminate the results by academic publishing. We would argue that for patients' self-tracking practices and insights to reach their full potential and result in changes to clinical practice, scientific publication and clinical validation is necessary.

Patients are beginning to report the results of their own N-of-1 studies in a wide variety of health conditions including diabetes type 1 [36], Crohn's disease [37], and food intolerance [38] and such reports are seen as a potentially important contributor to personalised clinical care [39]. Even though there are examples of patient driven smaller studies in PD [40,41] we were unable to find any scientific articles reporting specifically on patient-driven N-of-1 in PD. The lack of scientifically published reports does not necessarily mean that they do not exist. The first author has engaged in self-tracking to better understand the variations of her PD for a number of years and also writes about it on her personal blog (http://www.riggare.se). Her self-tracking work has been described in the popular press [42,43] but not in academic journals. She had been looking for an example suitable for an academic paper to demonstrate the usefulness of self-tracking for improving self-management and came across the work of one of the co-authors (Sturr) on social media. Sturr had been self-tracking her own PD and a collaboration was initiated.

Objectives

The objectives of the study were to: 1) explore the potential role for patient-driven N-of-1 studies as a tool for improving self-management in PD using the example of managing LID with nicotine, and 2) based on this example, identify some specific challenges of patient-driven N-of-1 studies.

METHODS

To meet the aims of the study, a placebo-controlled patient-driven N-of-1 study was designed using e-cigarettes to administer nicotine. The first author (Riggare) initiated the experiment and recruited other researchers, clinicians and patients to participate in conducting the study, including design, data collection, analysis, and reporting. The first author was also the subject of the study; she is an experienced self-tracker, had not smoked before and was on stable doses of PD medication at the time for the experiment. Subject characteristics are listed in **Table 3.1**.

Nicotine has been demonstrated as effective against LID in an animal model of PD (primates) without an increase in parkinsonian symptoms [44] and in a paper by Quik and colleagues [16] there is mention of a small trial on human subjects (phase I/II). Quik et al. states that four months of oral nicotine treatment in persons with PD with moderate disease decreased several measures of LID [16] but we have not been able to find any separate article reporting on the results from that trial.

TABLE 3.1: Subject characteristics.

Gender	Age (years)	Time since onset (years)	Time since diagnosis (years)	Type of PD	Hoehn &Yahr stage	PD medication incl daily dose
F	45	32	13	Juvenile onset PD (Parkin genetic form) with a 3 year history of falls and occasional LID	3	Levodopa/benserazid, 150 mg Entacapone, 1,000 mg Ropinerole, 10 mg Rasagiline, 1 mg Rivastigmine, 3 mg

Two identical sets of e-cigarettes (KangerTech mini starter kit) were purchased together with two bottles of e-juice of identical flavour, one with nicotine (3 mg/ ml) and the other without. The subject took additional levodopa (25 mg) an hour before the start of the experiment to increase the likelihood of dyskinesia. The e-cigarettes were used as therapeutical intervention.

In order to minimize exposure to LID, which is an uncomfortable and unwanted side effect, the experiment was conducted during as short a time span as possible. Based on prior experience by one of the authors (Sturr) with managing LID with e-cigarette, nicotine was expected to reduce LID within less than 30 seconds after administration of a few puffs. The subject took two to four puffs from the e-cigarette each time and took notes of her perception of the effect. Depending on the exploratory nature of the experiment, the number of puffs was not standardised. During the planning and undertaking of the experiment, notes were made to identify challenges specific to patient-driven N-of-1 studies.

Ethical considerations are an essential part of all research. Our study can be considered analogous to the self-experimentation performed by clinicians and researchers prior to testing interventions on the intended populations or healthy volunteers. Self-experimentation of that kind has previously been regarded as an essential part of ethical research [1]. Our study has not been reviewed by any ethical review board and there is support in the literature that N-of-1 studies often do not require IRB [45]. Nevertheless, ethical issues have been considered. The idea for the study came from the subject who was well-informed, knowledgeable and chose not only to participate voluntarily but also took a leading role in the planning, performing and analysis of the study. The potential risks for coercion or peer pressure were considered negligible. The subject was planning to conduct the experiment regardless of whether it was going to be submitted for publication or not. However, in order to share the results of the experiment as well as the experience of utilizing the patient-driven N-of-1 method

to a wider audience, it was considered ethical to publish the results in a scientific journal.

RESULTS

The experiment was conducted between 10 and 11 am local time on 23rd September 2016 in the Oregon Convention Centre during the 4th World Parkinson Congress. The assessments of the perceived effects of the e-cigarette by the subject together with notes taken by the subject are listed in **Table 3.2**.

During the first test (test A in **Table 3.2**), the subject could perceive no effect. At the start of test B however, a sense of calm spread through her body, leading to a reduction of dyskinesia. Furthermore, she experienced a clearness of mind that she had not felt in a long time. During tests C and D, the dyskinesia was also reduced but during test E the subject experienced no effect.

When the tests were unblinded, it was clear that the subject was able to distinguish a decrease of her LID from nicotine but no effect from placebo.

TABLE 3.2: Assessment of effects by the subject.

	Subject assessment	Subject notes	Key
Test A	Placebo	No effect	Placebo
Test B	Nicotine	Sense of calm spread through body, brain fog lifted	Nicotine
Test C	Nicotine	Less dyskinesia	Nicotine
Test D	Nicotine	Sense of calm, less dyskinesia	Nicotine
Test E	Placebo	No effect	Placebo
Correct (%)	100%		

During the planning, design, execution and analysis of this experiment a number of challenges specific to patient-driven N-of-1 studies were noted, which are listed below. The list is not intended to be exhaustive but gives some examples of considerations.

1. Planning. To develop the research idea, you have to make sure that the research you want to do fits the current state of research in that area. Hence, it is necessary to familiarize yourself with the research area in question, both state-of-the-art and some of the history in the area. To be able to do this, it is important to have access to published papers, also beyond open access articles. The research

question(-s) has to be developed and the study as a whole planned.
2. Research team. If you don't have all the necessary skills and knowledge yourself, a suitable research team has to be recruited. The team as a whole has to be able to plan, design, and conduct the study, as well as collect and analyse data.
3. Data collection. How can data best be collected and what are the best tools to use?
4. Analysis. How can the data best be interpreted? Are other experts needed to interpret the findings?
5. Publication of results. In order to publish the research, you have to conform to the guidelines and restrictions of scientific journals, which can be a challenge in itself. The cost for publishing may also be an issue as well as choosing the appropriate journal.

DISCUSSION

Our aim was to explore the potential role of patient-driven N-of-1 studies as a tool for improving self-management in PD by conducting an experiment using nicotine to reduce LID. The subject experienced a reduction in LID from nicotine and not from placebo which supports the notion that N-of-1 is potentially useful for enabling persons with PD to better understand and manage their condition. We also want to highlight that advanced technology was not necessary in order to achieve important insights.

Levodopa-induced Dyskinesia and Nicotine

LID is a potentially troublesome and common side effect of long term anti-parkinsonian treatment with significant negative impact on quality of life and few effective and available treatment options.

The e-cigarette was invented in 2003 and has since spread rapidly across the world. It consists of a battery, an atomizer and a reservoir containing the e-liquid. A heating coil inside the atomizer generates the aerosol. The e-liquid is available in a large number of flavours, both with and without nicotine. E-cigarettes can be a way to facilitate smoking cessation although health and safety are not fully understood and the long-term effects of inhaling vapours of nicotine and solvents are not currently known [46]. Further work is therefore needed in this area.

Benefits and Challenges of Patient-driven N-of-1

In PD, conventional N-of-1 studies have been used to explore effects of substances showing potential in primate studies [47,48]. We found two articles describing studies

of PD symptomology (e.g. LID) and in one case the effects on primates were also seen in human subjects [47]. An N-of-1 design has also been used to study the effect of espresso coffee on daytime somnolence in PD [49]. Espresso was considered efficacious compared to decaffeinated coffee in two of the four persons with PD included.

The use of patient-driven N-of-1 for PD should be further explored because persons with PD have relatively limited options for treatment, especially in the long term. Levodopa was first used to treat PD in 1961 [50] and remains the gold standard treatment 50+ years later, despite the common side effects. The highly individual aspects of PD also mean that it can be a challenge to find an optimal treatment regime and the progressiveness of the condition means that adjustments to the regime may be necessary also between clinical visits. N-of-1 studies offer a potential opportunity for persons with PD to be proactive in the management of their disease and learn more about their individual condition and treatments. The method can be used to generate hypotheses grounded in personal experience followed by testing on an individual basis. With access to methods to evaluate individual effects of various interventions, persons with PD may explore different treatments, both pharmacological and other available conventional and alternative interventions. If data and information can be collected in a structured way, this can also contribute to clinical research and practise. There is however more work needed on developing robust and scientifically sound methods for patient-driven N-of-1 studies.

Increasingly, patients of today are active in the management of their health and well-being, they find valuable health information online [51,52], connect with fellow patients in online communities [29] and use the information they find when communicating with healthcare providers [53]. Networks of patients connect online, test different ideas and share results, experiences and lessons learned. Social media enables patients to connect with and learn from other patients, clinicians and researchers all over the world. There are also scientific conferences that leverage this potential and one prominent example is the World Parkinson Congress (WPC), organised by the World Parkinson Coalition (http://www. worldpdcoalition.org), a non-profit organisation working to provide an international forum for knowledge and learning about PD actively engaging all stakeholders, physicians, scientists, nurses, rehabilitation specialists, caregivers and persons with PD. Our study originated from social media and was made possible through the work of WPC. The first author (Riggare) came across a video on social media of a fellow person with PD and co-author (Sturr) using nicotine distributed by means of an e-cigarette to manage her LID. Contact was made and the resulting collaboration led to conducting this study at WPC. We see that the combination of social media and conferences where patients can meet other patients as well as

clinicians and researchers has a strong potential for new findings and collaborations. The distinction we make between self-tracking and patient-driven N-of-1 has implications. Both have the benefit of drawing on motivation from the participant/researcher, something that has been identified as a key factor for success [54]. One main difference is that self-tracking can be conducted easily and can provide important insights without much planning or time for writing up the results. However, if you want to make your self-tracking publicly known via academic journals, according to our definition of patient-driven N-of-1, there are considerations to be made, as listed in the Results section. The main challenges of patient-driven N-of-1 studies were identified to be associated with planning of the study, recruiting a suitable research team, making sure the data collection is optimal, analysis of data, and publication of results. This process is of course not linear but rather iterative and explorative. For patients who are interested in performing N-of-1 studies, there are many hurdles to overcome in order to be able to share your results with the scientific community.

Limitations

Our study is not without limitations. Results from N-of-1 studies cannot be extrapolated beyond the subjects in the study. Results from multiple N-of-1 conducted in a standardised way can however generate results applicable to a wider population [45]. Another challenge is related to the demographics and effects of PD. Persons with PD are often older and may not be able to engage as actively in their self-management as our subject. The apathy that often is associated with PD can also make it difficult to expand the use of patient-driven N-of-1 into the wider population of persons with PD. However, for each person with PD that can be motivated to manage their condition more proactively, healthcare resources may be reallocated to those in more need of help. We therefore believe that the potential benefits justify further work in this area.

CONCLUSIONS

In conclusion, this study indicates that nicotine administered via e-cigarette may have an effect on levodopa-induced dyskinesia in individual patients with PD. The main contribution of this paper is however highlighting the work done by patients on a daily basis for understanding their conditions and conducting self-tracking experiments. These experiments and observations are rarely disseminated in academic journals in the form of patient-driven N-of-1 studies and therefore do not reach medical professionals in clinical practise in a validated manner. More work is needed to further develop methods for patient-driven N-of-1 studies for PD.

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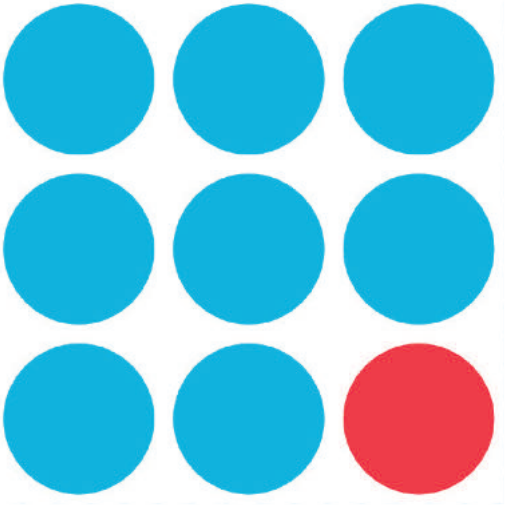
PART II

TRANSFERABILITY OF THE METHODS OF PERSONAL SCIENCE



What does it feel like to have PD? Naturally, I can only speak from my own experiences; I can't know what someone else's PD feels like. However, based on my interactions with the thousands of persons with PD that I've met over the years, I would say that the kinship I feel when meeting other persons with PD comes from a shared understanding of each other's struggles and challenges with our common "enemy". Even though we don't all have the characteristic tremor that is so strongly associated with PD, I think that we share a similar sense of frustration with the symptoms of PD and the side effects of our medications.

CHAPTER 4

Selfcare among persons with PD in Sweden

Published as: Patients are doing it for themselves: A survey on disease-specific knowledge acquisition among people with Parkinson's disease in Sweden.
Riggare S, Höglund PJ, Hvitfeldt Forsberg H, Eftimovska E, Svenningsson P, Hägglund M.
Health Informatics J. 2019 Mar;25(1):91-105.

ABSTRACT

Effective self-management is key to living well with Parkinson's disease and one important aspect is disease-specific knowledge. This article explores how people with Parkinson's disease in Sweden (1) acquire disease-specific knowledge and (2) use Parkinson's disease–related healthcare. Data were collected through an online survey, which had 346 respondents (16–87 years old, median age: 68 years, 51% male; time since diagnosis: 0–31 years, median time: 7 years). Our results show that disease-specific knowledge is mainly found online, especially for women with Parkinson's disease and persons with Parkinson's disease of working age, that most persons with Parkinson's disease in Sweden see their neurologist for 1 h or less per year and only one in two has regular contact with other Parkinson's disease–related healthcare professionals. We also find that people with Parkinson's disease reporting higher levels of specific knowledge also are more likely to be satisfied with the amount of time they get with their neurologist, regardless of the amount of time.

INTRODUCTION

Parkinson's disease (PD) is a chronic neurodegenerative disease resulting in motor and non-motor symptoms causing significant burdens on individual patients and family members, as well as on healthcare and society [1]. PD is extremely individual in nature and the range and fluctuations of symptoms often require complex medication regimens [2]. The prevalence increases with age; median age of onset is 60 years although 10 per cent of persons with PD are younger than 45 [1,3]. The number of persons with PD in Sweden is about 22,000 [4]. Worldwide, the number of persons with PD is predicted to double between the years 2005 and 2030 [5].

Healthcare has historically dealt with caring for acute injuries and illnesses where healthcare professionals were experts and patients passive recipients of care [6]. Chronic conditions, however, require a very different model for healthcare, one that is based more on patients' self-management and patient education [7,8].

When in need of health-related or medical information, people with chronic diseases turn to different sources. In a study from the United States [9], the following percentages were reported: health professionals: 93 per cent, friend or family member: 60 per cent, books or similar: 56 per cent, Internet: 44 per cent, insurance provider: 38 per cent, and other sources: 6 per cent. Although many studies explore patients' online information-seeking behaviour [10–13], we have not found any similar studies looking at other sources for other countries, including Sweden, or for persons with PD.

Internet access has accelerated the search for information and resources, and patients with chronic diseases actively use the Internet to search disease-related information outside of healthcare. For example, in the United States, 51 per cent of adults living with chronic conditions go online to find health-related information [9]. Another US study reports that about half of the population with chronic conditions would appreciate guidance when searching for health information online [14]. A survey study from Japan [15] demonstrated a lower use of Internet for health purposes; 23.4 per cent used a computer to acquire health information and 6 per cent used cell phones. We have not been able to find any similar study for a Swedish population but we know that Internet use in Sweden is high; in 2015, in total, 91 per cent of the population were online with slightly higher use (>95%) for ages 8–55 years [16]. There are, however, differences in use of the Internet across the population; a different study shows that among those 65 and older, being male, high education, not living alone, not being cognitively impaired, being younger within the studied population and urban living correlated significantly with higher Internet use [17].

Specifically in PD, effective self-management is crucial to successfully managing the disease and includes knowledge about the disease, medications and side effects, monitoring of symptoms, finding reliable sources of information and knowing when to take action [18–20]. A large majority of persons with PD want to be active in health-related decisions and to have access to correct and relevant information [21–23]. Online tools and services are frequently used among people with chronic diseases and have also been observed in PD [24–26]. A US study shows that persons with PD often have access to and feel comfortable using computers, mobile phones and the Internet [27].

The concept of health literacy has emerged as a way of describing, measuring and improving patient education. It was originally defined as 'the degree to which individuals have the capacity to obtain, process and understand basic health information and services needed to make appropriate health decisions' [28]. With the increasing use of online services, eHealth literacy has been developed. There are a number of different definitions of eHealth literacy and one of the most frequently used is 'the ability to seek, find, understand and appraise health information from electronic sources and apply knowledge gained to addressing or solving a health problem' [29]. It seems that health and eHealth literacy in the context of PD are under-researched concepts. A search using search terms 'health literacy' OR 'ehealth literacy' AND 'Parkinson*' in several databases (PubMed, Web of Science and CINAHL) revealed only three studies, two of which were abstracts for conference posters and the third, a pilot study. One of the conference abstracts determined the readability of letters sent from clinics to persons with PD and states that there might be a discrepancy between self-rated understanding by persons with PD and the actual readability [30]. The other one reports a study of health literacy in 121 persons with PD using two brief assessments and concluded that low health literacy is common in the investigated population and is likely to be even more prevalent in a general population of persons with PD [31]. The pilot study reports a prospective study of the functional health literacy of 44 men with PD and concludes that contrary to existing literature, persons with PD can be expected to preserve health literacy [32].

Rather than to study the full concept of health literacy, we wanted to study one specific aspect, namely, acquisition of disease-specific knowledge. This has previously been studied in the context of chronic disease [9,15], but we have not been able to find any previous studies of this aspect specifically for PD. In this article, we use the term disease-specific knowledge as meaning all knowledge relating to PD, including but not limited to knowledge about symptoms, medication and other treatments, side effects, disease management and healthcare provision. This is relevant because raising the level of disease-specific knowledge among persons with PD has been found to

increase health-related quality of life [33,34]. A recent study in China assessed the knowledge of PD among persons with PD and noted a great need for improvement in key areas such as disease management and awareness of medication side effects [35]. Since healthcare professionals and healthcare are traditionally the main sources of validated disease-specific knowledge [36], it is worth looking at time that is available to provide this knowledge. In the United States, persons with PD have appointments with their neurologist three to four times a year [37]; in Italy, one to three times [38], and in Sweden, once a year [4]. This indicates a discrepancy between the availability and significance of healthcare as a source of disease-specific knowledge.

We investigated two research questions:

- How do persons with PD in Sweden
    - Acquire disease-specific knowledge?
    - Use PD-specific healthcare?

METHODS

Data were collected from persons with PD in Sweden by means of a survey developed in a stepwise process. The survey (in Swedish) was designed and distributed using the online tool Typeform [39].

General information about the survey (purpose of the study, investigator, instructions for responding) was included and questions were kept short and focussed to reduce the risk of respondents abandoning the survey before completion [40]. The survey questions are listed in Table 3 of Appendix 1 and include questions on background (gender, year of birth, place of living and education level), year of diagnosis and the importance, level and main source of disease-specific knowledge. There were also questions on how much time is spent in healthcare every year (neurologist and other healthcare professionals), as well as an assessment of time sufficiency. Response options were numerical, categorical or free text. When asked about their opinion, respondents were given a five-point Likert scale to choose from, where the middle option signified a neutral opinion.

The survey was first tested on a smaller pilot group with PD, four participants (50% women) with varying ages (49–67 years) and time since diagnosis (6–13 years), in a fully functional online form. Some minor text edits were made before distributing more broadly using the online tool. To maximise the number of responses, the web link to the survey was distributed to persons with PD in Sweden via email to patient organisations and patient groups, as well as social media and personal networks. No

incentives were offered for responding. The survey was made available as soon as it was ready and data collection was terminated when the number of new responses tapered off. It was online for 4 weeks (7 March–4 April 2015), after which the results were downloaded and analysed.

Only respondents living in Sweden were included in the analysis and duplicate answers were excluded. Age and time since diagnosis were calculated from year of birth and year of diagnosis. Categorical and numerical variables were analysed with an interactive calculation tool [41] using the $\chi 2$ test with statistical significance defined at $p < .05$.

TABLE 4.1: Respondent characteristics.

Respondent characteristics	Number of respondents	Interval	Median	
Age	346	16-87 years	68 years	51% male
Time since diagnosis	335	0-31 years	7 years	
Education level				
Compulsory school (<9 years)	74 (21%)			
Upper secondary school (9-12 years)	93 (27%)			
University (>12 years)	179 (52%)			

This study is exempted from ethical approval by the regional ethical review board in Stockholm (according to decision 2015/1572-31/4).

RESULTS

Background data

The survey had 346 valid responses, 11 of which did not give year of diagnosis, and 48 per cent of the unique visitors completed the survey. The age (**Table 4.1** and **Figure 4.1**) and gender (**Table 4.1** and **Figure 4.2**) distributions are consistent with what would be expected in a population with PD and representative for Swedish persons with PD compared to a study by Lökk et al [4]. There is one 16-year-old respondent, who is unusually young for PD but rare forms have been reported with juvenile onset [42].

Our respondents are relatively well educated (**Table 4.1**), 52 per cent have completed more than 12 years of education, which is more than the general population in Sweden, where 30–35 per cent of the population are reported to have studied for 12 years or more [43].

Disease-specific knowledge

Results regarding self-reported importance, level and main source of disease-specific

knowledge are presented in **Table 4.2**. In total, 91 per cent of the respondents considered knowledge about PD important (4 or 5 on the Likert scale for *Importance of knowledge*) and 55 per cent reported to have been able to acquire the knowledge they need (4 or 5 on the Likert scale for *Level of knowledge*). When asked which is their main source of disease-specific knowledge, 36 per cent responded that they had found the information themselves online, 29 per cent from patient organisations and similar, and 25 per cent from healthcare.

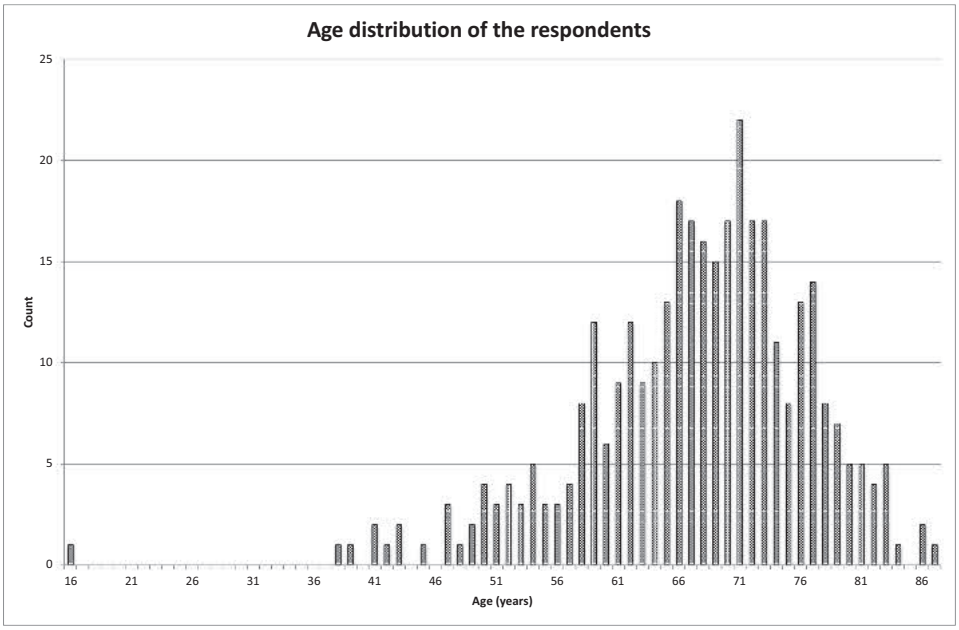


FIGURE 4.1: Age distribution of the respondents.

People with different education levels could be expected to report different levels and sources of knowledge; however, chi-square analyses of our respondents showed that neither level (p = .58) nor source of knowledge (p = .18) are significantly associated with education level. Furthermore, age, gender or time since diagnosis could be expected to influence level and source of knowledge. We have chosen to use the definition of 'older adults' from the Swedish National Board for Health and Welfare [44] (65 years). For 'time since diagnosis, we use the median value (7 years) for separating into two groups. We found that the self-reported level of knowledge is not significantly associated with age (p = .41), gender (p = .64) or time since diagnosis (p = .41). Similarly, the self-reported main source of knowledge is not significantly associated with time since diagnosis (p=.18).

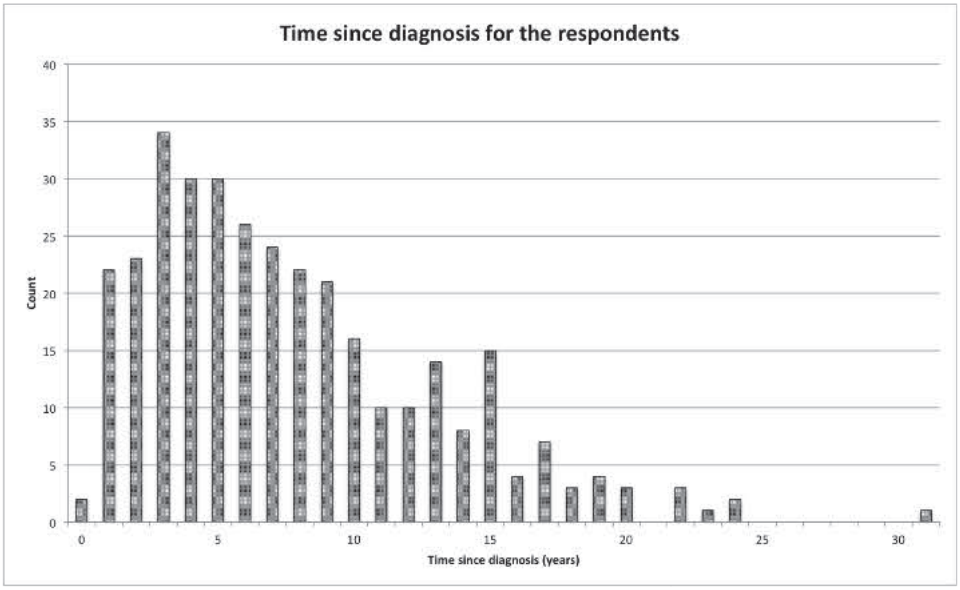


FIGURE 4.2: Time since diagnosis for the respondents.

TABLE 4.2: Importance, level, and source of knowledge.

Importance of knowledge	*1*	*2*	*3*	*4*	*5*	*Total*
How important is knowledge about your disease to you? (1=unimportant, 5=very important)	0	9	22	53	262	346
	0%	3%	6%	15%	76%	100%
	1	*2*	*3*	*4*	*5*	*Total*
Level of knowledge						
Have you been able to acquire the knowledge you need about your disease? (1=not at all, 5=absolutely)	17	35	103	121	70	346
	5%	10%	30%	35%	20%	100%
Source of knowledge						
Which of these is your main source of knowledge about your disease? Pick one.						
Information I have found myself online	123	36%				
Information from patient organisations etc	100	29%				
Information from healthcare	87	25%				
Information from other patients	18	5%				
Information from family, relatives and friends	9	3%				
Other sources	9	3%				
Total	346	100%				

Analyses show, however, that significantly different main sources of knowledge are reported depending on age ($\chi 2 = 13.6$, df = 3, p = .003) and gender ($\chi 2 = 9.62$, df = 3, p = .022). Online information is the most important main source of disease-specific knowledge for persons with PD under 65 years of age, whereas the group 65 and older more often report patient organisations as their main source (**Figure 4.3**). It is worth noting that the 31 per cent of the group 65 and older who report Internet as their main source of disease-specific knowledge is still a relatively high number. Men and

women also report significantly different sources (**Figure 4.4**); women more often find their information online. To enable the use of chi-square testing for analysing source of knowledge, the response options '*other patients*', '*family, relatives and friends*' and '*other sources*' were merged.

Time in healthcare

When it comes to time spent in healthcare (**Table 4.4** of **Appendix 4.1**), 35 per cent visited their neurologist once and 38 per cent twice during 2014. Three visits were made by 10 per cent of the respondents and 9 per cent had four visits or more. This means that 8 per cent (n=29) of our respondents did not see a neurologist at all during the year. As for the length of visits, 14 per cent met with their neurologist for up to 15 min, 48 per cent between 15 and 30 min, 23 per cent for 30–45 min and 14 per cent for an hour or more per visit. We calculated the total yearly time with the neurologist from number of visits and time per visit. In total, 60 per cent (n=206) saw their neurologist for up to an hour during the year (**Figure 4.5**; **Table 4.4** of **Appendix 4.1**).

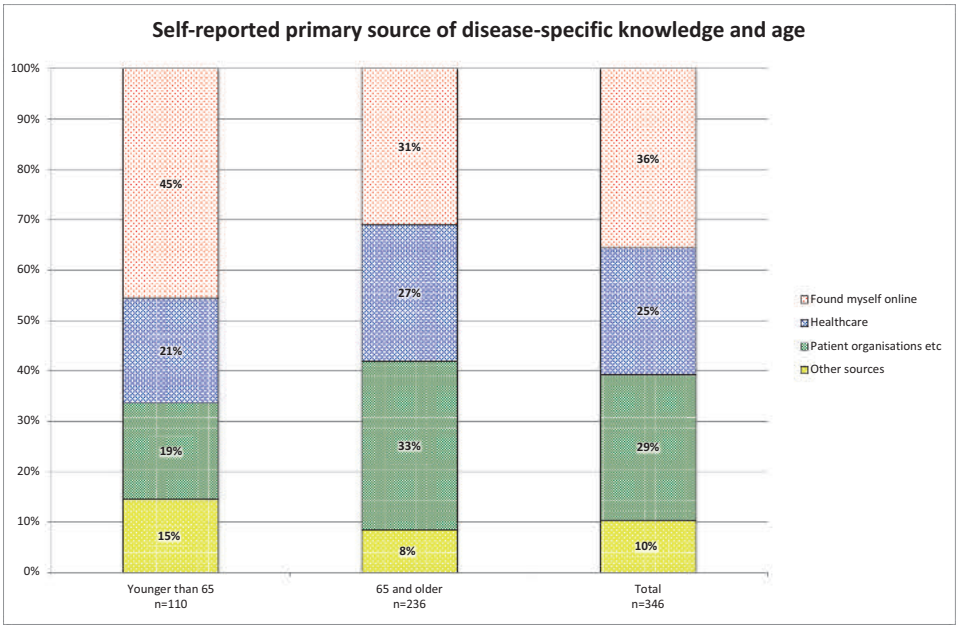


FIGURE 4.3: Self-reported primary source of disease-specific knowledge for different age groups.

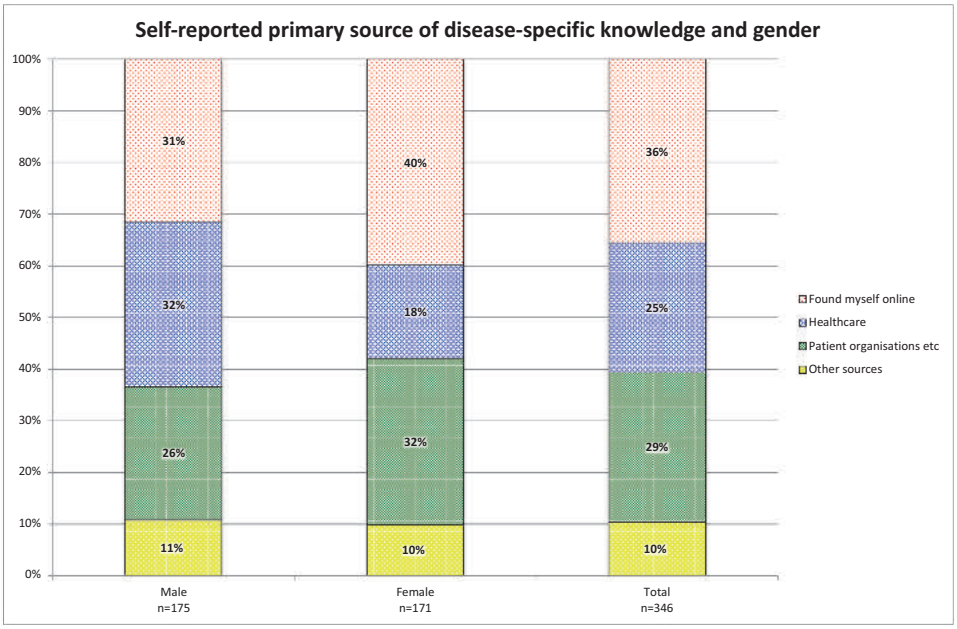


FIGURE 4.4: Self-reported primary source of disease-specific knowledge for different genders.

When asked if they considered the time they have with their neurologist to be sufficient, 35 per cent of the respondents said no (1 or 2 on the Likert scale), 43 per cent said yes (4 or 5 on the Likert scale) and 22 per cent were neutral (3 on the Likert scale) (**Table 4.4** of **Appendix 4.1**). As expected, there is a significant association between spending more time per year with your neurologist and being satisfied with the amount of time spent ($\chi 2 = 16.8$, df= 4, p = .002).

On the question about regular contact with other healthcare professionals, 47 per cent (n = 161) reported that they had met with a nurse, speech therapist, physiotherapist or similar during 2014 (**Table 4.5** of **Appendix 4.1**). About one-third of the people with no time with their neurologist (9 out of 29) reported that they had regular contact with other healthcare professionals, for example, nurse, physiotherapist or speech therapist. Of these, one person had 10–20 h and the remaining eight had up to 5 h during the year.

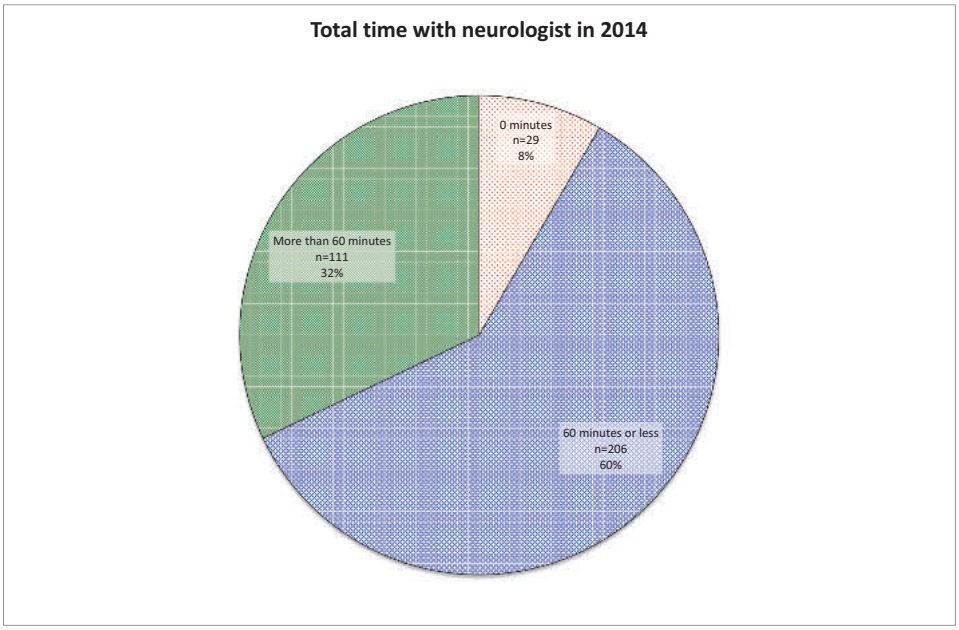


FIGURE 4.5: Total time with neurologist in 2014.

One could expect that people who spend more time with their neurologist per year would report higher levels of disease-specific knowledge and also more often report healthcare as their main source of knowledge. Chi-square analyses show, however, that yearly time with neurologist is not significantly associated with level ($p = .43$) or main source of knowledge ($p = .15$).

A person's gender, level of education or age could be expected to have an effect on their expectations on and satisfaction with healthcare. Furthermore, that the time since diagnosis might influence how much contact with the neurologist is perceived as needed. Analyses of our data show, however, that satisfaction among persons with PD of the time they get with their neurologist is not significantly associated with age ($p = .17$), gender ($p = .63$), time since diagnosis ($p = .44$) or education level ($p = .83$). Furthermore, we also found a significant association between self-reported level of knowledge and satisfaction with the time with the neurologist every year ($\chi^2=30.1$, $df= 4$, $p<.001$). This means that persons with PD reporting a higher level of knowledge were more likely to be satisfied with the time they had with their neurologist, regardless of how short or long that time was.

DISCUSSION

The aim of this study was to explore how persons with PD in Sweden acquire disease-specific knowledge and to investigate the use of PD-related healthcare in Sweden. The results from our survey indicate that Swedish persons with PD mainly acquire their disease-specific knowledge online and that the Internet is an even more important source of information for women with PD, as well as persons with PD under 65 years of age. We also see that most persons with PD in Sweden see their neurologist for 1 h or less per year. In the following sections, the results and methods will be discussed in more detail.

Discussion of results

It is hardly a surprising finding that the Internet is an important source of knowledge for persons with PD considering the increasing online presence of patients across all diseases. Age and gender seem to have a significant influence on the main source of knowledge, influence that appears independent from time since diagnosis, education level or time spent with neurologist per year. The fact that our data show that women to a higher degree find their information online could be seen as contradicting earlier research that suggests that men in Sweden are more online than women [17] but could also be seen as an indication that even though women are less online than men, they mainly find their disease-specific knowledge online.

Should the fact that people find their disease-specific information online be perceived as a problem? Not necessarily, because although it is important to acknowledge that it can be difficult finding correct and relevant information online, Internet does enable patients to more effectively manage their health and healthcare [45]. 'Internet-informed' patients influence the patient–provider relationship and contribute to making healthcare more patient-centred while healthcare professionals can overestimate the risks of online health information [46,47]. Results from a study among American patients with chronic conditions showed that a majority, 94 per cent, reported that they had not been harmed by health information they found online [9]. It is, however, important to note that Internet use can be an issue for persons with PD. Excessive use of Internet may be a sign of impulse control dysfunction, a known side effects of PD medications [48,49].

The effects of PD can also have an influence on our results. With the progression of the disease, for example, hand function may be affected, presenting as impaired fine motor skills and/or severe tremor. This could result in reduced ability to use keyboard, computer mouse or touch screens, hence making it more difficult to respond to online surveys.

The quality and accessibility of the information provided online can potentially be an issue. Indeed, the quality of information found can be difficult to assess for persons with PD. Misinformation can be both deliberate, with the purpose to promote a specific product or cause, and unintended mistakes. Both of these can be problematic and should be explored further. When it comes to readability, a study of PD information websites in English aimed towards persons with PD showed that the majority of the sites studied did not comply with readability guidelines [50]. Whether or not this is the case for Swedish websites with information on PD is not known, and based on the survey results, we cannot know which sites the respondents use to gather information, nor if our respondents defined 'the knowledge you need' in the same way or regarded the information they found as being credible, reliable and helpful. In accordance with existing research, we also cannot know in what way the information given is perceived and understood [8].

Would it not be better if healthcare took a larger responsibility in educating patients? There may of course be a role here for healthcare professionals to involve persons with PD more in their treatment and care, and according to a Dutch study, persons with PD expect healthcare to provide relevant information, tailored to the individual's needs [22]. However, as is reflected in a different study, advances in medicine and time constraints in healthcare make it hard for healthcare professionals to keep up with new knowledge and patients' needs and expectations, which often leaves patients feeling frustrated with the information provided [45]. The study even suggests that patients who are more Internet-savvy than their providers often feel better able to find the health-related information they need by themselves online [45]. As we can see from our results, most persons with PD have 60 min or less with healthcare annually, leaving little room for continuous information provision or patients' questions. In combination with the high Internet use in Sweden, even among the older population, it could be assumed that Swedish persons with PD might sometimes be able to find the information they need more easily than their healthcare providers.

It is also important to note that there is a difference between information and knowledge. Healthcare professionals might feel confident that they provide the right information at the right time but, for time constraints or other reasons, they are not able to ensure that the information is properly received and transformed into knowledge by the individual. The perception of what kind of information is relevant at different times and stages of the disease might also vary between healthcare and persons with PD, and even between persons with PD. This makes it, of course, very difficult for healthcare professionals to provide information relevant to each person with PD at every occasion. Furthermore, we would argue that different sources might provide different types of information. It might be feasible to think that certain types of

information would be best if given by healthcare, whereas other kinds might be best found elsewhere, a topic that would need further exploration in the future.

We also see the fact that persons with PD find their information online as part of the on-going shift from patients being passive recipients of care to active participants who have the possibility to be experts in managing their own disease and situation [23]. Considering the complexity of the disease and treatments, we believe it is unreasonable to expect the limited time persons with PD have in healthcare to be sufficient to adequately address all the relevant issues. We therefore propose that other avenues for supporting the acquisition of disease-specific knowledge for persons with PD are explored. Our data show that also fellow patients, in the form of patient organisations and similar, are important sources of disease-specific knowledge. We propose to combine the power of the Internet with the force of patients to complement the current information provision by healthcare. By utilising the networking powers of online communities and online learning, we believe that some of the pressure on healthcare can be alleviated. This is developed further in 'Future work' below.

Discussion of methods

Choosing an online survey as a data collection method has its own advantages and limitations. Although online surveys are a fast and efficient way to collect data [40], they bias the results in favour of people who are already active online and probably use Internet as a knowledge source. Our respondents are more educated than the Swedish population in general and a higher level of education has been shown to significantly predict the use of Internet for health purposes [45]. This means that our results might overestimate the proportion of people who mainly find their information online. However, since Sweden, in general, has a very high use of Internet, the overestimate is likely to be minor.

Respondents were asked to self-assess their level of disease-specific knowledge and relate it to the knowledge they considered themselves needing. They were also asked to self-report the frequency and length of healthcare visits. Relying on self-assessments and recall gives rise to uncertainties in the responses.

Despite the weaknesses described above, our results contribute to new knowledge in an underexplored research field, and they point towards a development where the importance in online sources for patients' knowledge acquisition increases.

Our survey collected responses from about 1.5 per cent of all persons with PD in Sweden and when comparing with a study reporting age and gender distribution in that population [4], our population seems reasonably representative, with some lack

of responses in the higher age groups.

Future work

As far as the authors know, this is the first study exploring acquisition of disease-specific knowledge by persons with PD. Our questions were not detailed but rather general. Future research should focus on exploring the satisfaction of persons with PD with online information, investigating whether different sources provide different kind of information and individualising the knowledge type, form and delivery to patient needs and preferences. Exploring ways to objectively assess the disease-specific knowledge of persons with PD is another future research focus, as well as looking at potential methods for ensuring the quality of online health information, to avoid inappropriate guidelines and recommendations. In a subsequent next step, the results from the proposed work, an online service for implementing these ideas could be developed.

CONCLUSION

Persons with PD in Sweden mainly find their disease-specific information online, especially so for women and people in working age. At the same time, healthcare has a very limited possibility to provide disease-specific information since most persons with PD see their neurologist for 1 h or less per year. Moreover, only 1 out of 2 has contact with other PD-specific healthcare professionals. Given the overburdened healthcare system, it would be worth exploring other ways of guiding patients to relevant and accurate health information, such as education programmes and peer support networks delivered by patient organisations or other entities independently from healthcare. The timing, content and delivery mode of PD-specific knowledge need to be addressed in future research.

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APPENDIX 4.1

TABLE 4.3: Survey questions and response options.

Question	*Reply options*
Gender?	M/F
Year of birth?	Numeric
County council of residence?	Dropdown of all Swedish county councils
Highest completed education?	Compulsory school (<9 years)
	Upper secondary school (9-12 years)
	University (>12 years)
Year of diagnosis?	Numeric
How important is knowledge about your disease to you?	Scale from 1 to 5 where 1=unimportant, 5=very important
Which of these is your main source of knowledge about your disease? Pick one.	Healthcare
	Patient organisations and similar
	Found myself online
	Other patients
	Family, relatives and friends
	Other sources
Have you been able to acquire the knowledge you need about your disease?	Scale from 1 to 5 where 1=not at all, 5=absolutely
How many times did you see your neurologist during 2014?	None
	Once
	Twice
	Three times
	Four times or more
	I do not have a neurologist
Non-mandatory comment about visits to the neurologist	Free text
How long is each visit on average? Pick the time closest to the actual time.	About 15 minutes
	About 30 minutes
	About 45 minutes
	An hour or more
Do you also have regular contacts with other healthcare professionals, for example nurse, speech therapist, physical therapist, social worker?	Yes
	No
	I do not know
Total time spent in healthcare for PD during 2014 (neurologist visits excluded)	Up to 5 hours
	Between 5 and 10 hours
	Between 10 and 20 hours
	More than 20 hours

TABLE 4.3: Continued

Question	*Reply options*
Non-mandatory comment about your other healthcare contacts	Free text
Do you think your time with the neurologist is sufficient?	Scale from 1 to 5 where 1=not at all, 5=absolutely
Non-mandatory comment about the time spent with the neurologist	Free text

TABLE 4.4: Time with neurologist.

Number of visits							
How many times did you see your neurologist during 2014?	None	Once	Twice	Three times	Four times or more	I don't have a neurologist	Total
	25	120	133	34	30	4	346
	7%	35%	38%	10%	9%	1%	100%

Length per visit						
How long is each visit on average?	About 15 mins	15-30 mins	30-45 mins	1 hour or more		Total
	50	165	81	50		346
	14%	48%	23%	14%		100%

Total time						
Total time with neurologist in 2014	0 mins	15-60 mins	61-120 mins	>120		Total
	29	206	92	19		346
	8%	60%	27%	5%		100%

Time sufficiency						
	1	2	3	4	5	Total
Do you think your time with the neurologist is sufficient? (1=not at all, 5=absolutely)	67	55	76	72	76	346
	19%	16%	22%	21%	22%	100%

TABLE 4.5: Total time spent in healthcare for PD.

Total time spent in healthcare for PD during 2014 (neurologist visits excluded)	Count
<5 hours	70
5-10 hours	18
10-20 hours	26
>20 hours	47
TOTAL	161

NOTE

As is pointed out in **Chapter 1**, personal science is an emerging research field that can be seen as a new kind of single subject research design. The fact that the field is new means that when the study reported in this chapter was conducted, slightly different terminology was used.

In this chapter, the term *self-tracking* can mean both *personal science* and the associated method for data collection.

CHAPTER 5

Personal science among persons with PD in Sweden

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BMC Med Inform Decis Mak. 2019 Aug 30;19(1):175.

ABSTRACT

Background

This study explores opinions and experiences of persons with Parkinson's disease in Sweden of using self-tracking. Parkinson's disease (PD) is a neurodegenerative condition entailing varied and changing symptoms and side effects that can be a challenge to manage optimally. Patients' self-tracking has demonstrated potential in other diseases, but we know little about PD self-tracking. The aim of this study was therefore to explore the opinions and experiences of persons with PD in Sweden of using self-tracking for PD.

Method

A mixed methods approach was used, combining qualitative data from seven interviews with quantitative data from a survey to formulate a model for self-tracking in PD. In total 280 persons with PD responded to the survey, 64% (n = 180) of which had experience from self-tracking.

Result

We propose a model for self-tracking in PD which share distinctive characteristics with the Plan-Do-Study-Act (PDSA) cycle for healthcare improvement. Persons with PD think that tracking takes a lot of work and the right individual balance between burdens and benefits needs to be found. Some strategies have been identified here; to focus on positive aspects rather than negative, to find better solutions for their selfcare, and to increase the benefits through improved tools and increased use of self-tracking results in the dialogue with healthcare.

Conclusion

The main identified benefits are that self-tracking gives persons with PD a deeper understanding of their own specific manifestations of PD and contributes to a more effective decision making regarding their own selfcare. The process of self-tracking also enables persons with PD to be more active in communicating with healthcare. Tracking takes a lot of work and there is a need to find the right balance between burdens and benefits.

BACKGROUND

Parkinson's disease (PD) is a neurodegenerative condition associated with a wide variety of motor and nonmotor symptoms. The manifestations of the disease as well as treatment regimens are often highly individual in nature, making the condition a challenge to manage optimally [1]. PD is primarily diagnosed on the basis of the four cardinal symptoms: tremor, bradykinesia, rigidity and problems with balance and gait. In addition, there are a number of potential other symptoms and medication side effects, related to motor and non-motor functions and the condition can significantly affect quality of life [1,2]. Median age of onset is 60 years and prevalence increases with age, the incidence between 70 and 79 years of age is 93.1 per 100,000 person years [2]. Medical treatment for PD relies primarily upon oral medications, often with a complicated regimen to follow along with the added risk of side effects. Types, amounts, and combinations of medication prescribed varies with national prescribing patterns [3], age of onset and duration disease [4], and use of brain surgery [5], but typically the number of tablets increases with duration of disease. A survey study of persons with PD in the US, France, Germany, Italy, Spain, and the UK found a mean daily intake of about 3 tablets for early stage and about 9 tablets for persons with advanced stage disease [6]. In advanced disease, brain surgery, such as Deep Brain Stimulation (DBS) and infusion therapy, such as duodopa and apomorhine are also used [1]. The effects of PD as well as the medical treatment are often of a very fluctuating nature and symptoms can differ from one minute to the next [1,2]. In a previous survey among persons with PD performed in Sweden, a majority of respondents saw their neurologist for one hour per year or less [7][4].

Across diseases, patients' self-tracking of symptoms is an area of increasing interest in healthcare and research. For many diseases, self-tracking of biomarkers and symptoms have been demonstrated to improve disease management [8] and clinical outcomes [9].

Self-tracking can be supported by the use of technology and different technical solutions for clinical use is an area of substantial interest in PD. The views of clinicians of the uses of technology seem to differ somewhat from that of persons with PD. Clinicians expressed that technology is most valuable when it can be related to an effective therapy [10], meaning mainly motor symptoms. Furthermore, that the main reasons for using technology are 1) research and 2) evidence-based medicine and clinical care [11]. Persons with PD consider motor and non-motor symptoms equally important [12] and expect technology to be able to capture the full complexity of PD [13]. A Swedish focus group study of the potential benefits and barriers of using wearable technology for PD and epilepsy found that persons with PD saw a potential

4 Chapter 4 of this thesis

to use technology for their own self-management including adjustment of medication [14]. Healthcare professionals in the same study expressed concerns about persons with PD adjusting their medication based on tools not provided from the clinic.

The temporal variability of PD symptoms in combination with the limited time persons with PD have with their clinicians implies a substantial potential for improvement with the use of self-tracking to achieve a better understanding of the condition. Previous studies have focused on clinical aspects [15] and hypothetical discussions [14]. To the best of our knowledge, the only previous study investigating the practical patient perspective on self-tracking in PD is a case study by two of the four authors to this study [16][5]. No studies have been reported looking at a larger group of persons with PD and their self-tracking practices, to see if there is a potential for a better understanding regarding their condition. What contribution can access to more objective data about one's own symptoms and health make? The aim of this study was therefore to explore the opinions and experiences of persons with PD in Sweden of using self-tracking for PD with a focus on variety of experiences rather than representativity.

METHOD

To identify the opinions and experiences of persons with PD regarding self-tracking, a mixed method approach and triangulation were used, to combine qualitative data from interviews with quantitative data from a survey. A mixed methods research approach can be suitable when aiming to explore a research question in both breadth and depth [17].

Study design

We chose to combine data from interviews with results from a survey. To increase our understanding for self-tracking in the context of PD, we first conducted in-depth interviews with persons with PD (n = 7) with experience of performing self-tracking. The results of these interviews then informed the design of a survey distributed more widely in the Swedish PD community. Analysis of both interviews and survey results informed the design of a model for self-tracking in PD. An overview of the study design is presented in **Figure 5.1**.

5 Chapter 2 of this thesis

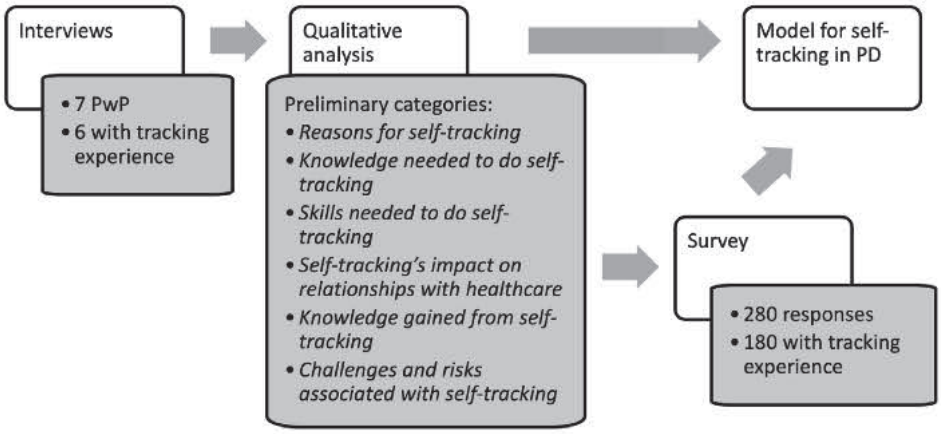

FIGURE 5.1: Study design.

Qualitative data collection

We used a purposive sampling strategy [18] informed by one of the authors (SR), recruiting participants from personal networks and contacts from previous work. We wanted to elicit the views and opinions of persons with PD on self-tracking and were looking specifically for persons with personal experience of self-tracking. Our respondents had all expressed an interest in self-tracking and all except one had personal experience of self-tracking. In order to obtain a broad perspective, efforts were made to find participants with varying backgrounds, ages, geographic location, and PD characteristics (current treatment, severity of disease). The participants all have PD without cognitive decline. **Table 5.1** gives an overview of the participants.

A semi-structured interview guide was developed specifically for this study, containing questions related to information on background, disease characteristics (time of diagnosis, symptoms), interactions with healthcare relating to PD, and self-tracking (see interview guide as **Appendix 5.1**). The interview guide was pilot tested with three persons with PD and minor adjustments were made prior to the first interview. Interviews with persons with PD other than the ones from the pilot test were conducted between October 2015 and August 2016. The duration of all the interviews was 283 min.

The first author, who is part of the PD community, conducted the interviews with the help of another person with PD using an iterative approach. After each two-three interviews, a preliminary analysis was performed to evaluate the level of saturation [19]. The interviews were continued until saturation was reached related to the topic of self-tracking.

During the interviews, background information (age, time since diagnosis, current treatment etc) was collected, see **Table 5.1**.

TABLE 5.1: Characteristics of interview respondents (n=7).

Gender (men/women), *n*	3/4
Age (years), median (min/max)	58 (52/67)
Years since diagnosis, median (min/max)	5 (3/13)
Current PD treatment, (oral/advanced)	(7/2*)
Personal experience of self-tracking, *n* (N)	6 (7)

* one DBS, one Duodopa

Qualitative analysis

The interviews were recorded, transcribed verbatim and analysed qualitatively using inductive qualitative content analysis [20,21]. The analysis was conducted in Swedish in order to stay as true as possible to the meaning of the text [22]. Two of the authors (SR and TSD) listened to the recordings, read the transcripts and sorted the text into two content areas: self-tracking and collecting data in collaboration with healthcare. SR and TSD then independently selected relevant data into one text (the unit of analysis). The data selection was verified among all four authors. In the next phase, the text was divided into condensed meaning units that were abstracted and labelled with codes. The codes as a whole were compared and organised in preliminary sub-categories and categories, representing the manifest content of the data. All four authors met several times to compare and discuss the codes, sub-categories, and categories. Where opinions varied, the cases were discussed until consensus was achieved. In the final phase of the analysis, in order to increase credibility, illustrative quotes were selected and translated to exemplify each category. The translations were verified by all authors.

The qualitative analysis of the interviews resulted in six distinct categories, namely *Reasons for self-tracking*, *Knowledge needed to do self-tracking*, *Skills needed to do self-tracking*, *Self-tracking's impact on relationships with healthcare*, *Knowledge gained from self-tracking*, and *Challenges and risks associated with self-tracking* (see also **Figure 5.1**). These preliminary categories were used to design the survey.

Quantitative data collection

Based on the categories from the qualitative analysis of the interviews, a survey was developed in Swedish, specifically for this study (see the survey as **Appendix 5.2**). Since

91% of the Swedish population uses the internet [23], an online survey was considered appropriate. The survey was designed and distributed using Google Forms. General information about the survey (purpose of the study, investigator, and instructions for responding) was included. The survey comprised six sections; background, experience of self-tracking, reasons for self-tracking, approach and use of self-tracking, self-tracking's influence on relationships with healthcare, and challenges and risks associated with self-tracking. Responses included multiple-choice options (with both "check only one" and "check all that apply") and a five-tiered Likert scale (with options *strongly disagree*, *disagree somewhat*, *neither agree nor disagree*, *agree somewhat*, and *strongly agree*). Questions were kept short and focussed to reduce the risk of respondents abandoning the survey before completion [24].

The link to the survey was distributed via patient organisations, social media and personal networks. Since our focus was on variety rather than representativity, we prioritised a wide reach over being able to calculate response rate. Data were collected between December 7, 2017 and January 7, 2018.

Quantitative analysis

For the purpose of analysis, the five-tiered rating was replaced by the options *disagree* (including the options *strongly disagree* and *disagree somewhat*), *neutral*, *agree* (including the options *strongly agree* and *agree somewhat*). An online calculation tool [25] was used for statistical analyses using the $\chi 2$ test with statistical significance defined at $p < .05$.

RESULTS

The results from the interviews and the survey were combined to create a conceptual model describing our understanding of self-tracking in PD. Analysis of the qualitative and quantitative data resulted in five distinctive categories, which are further described in the following sections. The categories are: *Why I self-track, How and what I self-track, Lessons learned from self-tracking, Risks related to self-tracking,* and *Self-tracking and healthcare*.

Background data

In total, the survey had 280 complete responses from Swedish persons with PD whereof 180 had experience from self-tracking. The characteristics of the respondents, including age, time since diagnosis and education level are given in **Table 5.2** (with self-tracking experience).

TABLE 5.2: Characteristics of survey respondents with self-tracking experience (n=180).

Age	26-35 yrs	36-45 yrs	46-55 yrs	56-65 yrs	66-75 yrs	76-85 yrs	>86 yrs
Women	1	6	14	28	30	9	
Men			11	23	46	11	1
%	1%	3%	14%	28%	42%	11%	1%
Time since diagnosis		**<1 yr**	**1-5 yrs**	**6-10 yrs**	**11-15 yrs**	**16-20 yrs**	**>20 yrs**
Women		5	38	25	17	2	1
Men		1	31	32	17	8	3
%		3%	38%	32%	19%	6%	2%

Highest completed education level	Compulsory school (<9 yrs)	Upper secondary school (9–12 yrs)	University (>12 yrs)
Women	5	28	55
Men	10	25	57
%	8%	30%	62%

Regardless of self-tracking experience or not, the respondents were similar in age distribution, education levels and proportion male/female. There was, however, a significant difference regarding time since diagnosis (see **Figure 5.2**); survey respondents who had been diagnosed more than five years were more likely to have tried self-tracking (71%) than persons with PD diagnosed five years or shorter (57% have tried self-tracking) ($\chi 2(2) = 6.066$, $p = .014$).

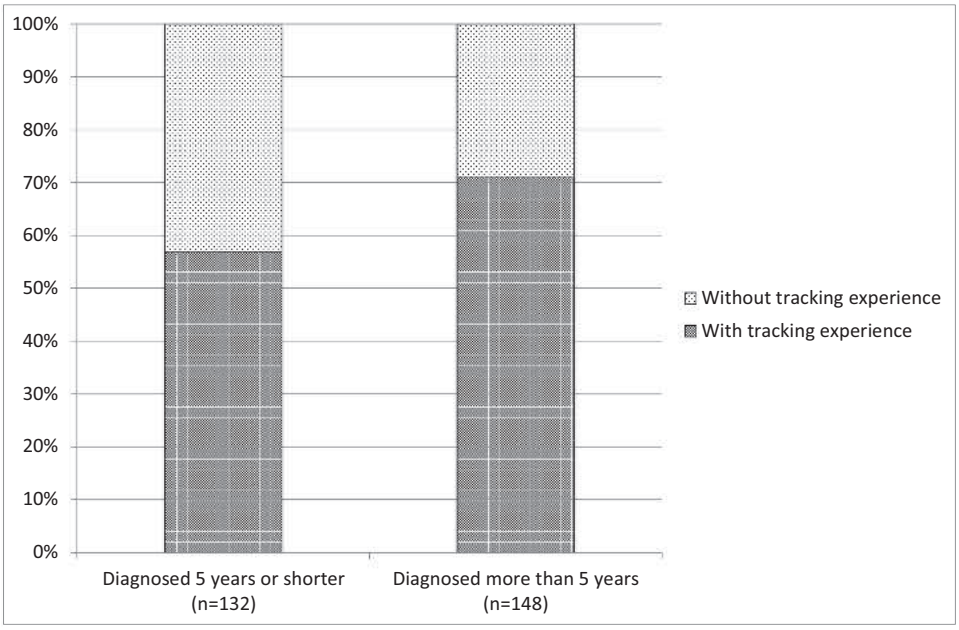

FIGURE 5.2: Tracking experience according to time since diagnosis.

To meet the aim of the study, the analyses and construction of the model were made using the survey results from persons with PD with self-tracking experience (n = 180). Of those respondents 49% were women, average age was 64.4 years and average time since diagnosis was 7.7 years.

Why I self-track

All respondents in the interviews talked about reasons for self-tracking. Several of them mentioned that they have a mind-set for self-tracking and therefore may be more interested than persons with PD in general. They also demonstrated an awareness of the highly individual presentation of PD, that the illness will be very different for different individuals and that each person needs to understand their own symptoms and how to manage them. Some respondents also described self-tracking as a means to achieve increased awareness of their illness and its progression.

> *R3: "I expect tracking to help me more clearly see how my disease really is, now it's mostly guesswork."*

Similarly, in the survey (see **Figure 5.3**), the most common response to why people self-track, was that they expect it to enable them to understand their PD better (74%). Persons with PD younger than 65 are significantly more likely to state that self-tracking enables them to understand their PD better (87%) than persons with PD older than 65 (63%) ($\chi 2(2) = 13.215$, $p = .001$), see **Figure 5.4**.

The interview respondents also mentioned self-tracking as a tool to find and understand correlations between health status or symptoms and medication intakes. It is a way to recall how they were doing over time, for example to remember how symptoms fluctuate. Both of these reasons were chosen by 73% of the respondents of the survey, see **Figure 5.3**.

Some of the interview respondents talked about how they take an active approach in the management of their PD. Self-tracking was described as a way to stay in control, and to take personal responsibility for one's health using an active approach. In the survey, 71% stated that tracking enabled them to take an active approach in the management of their PD, see **Figure 5.3**. Persons with PD younger than 65 were also significantly more likely to take an active approach (83%) than persons with PD older than 65 (60%) ($\chi 2(2) = 12.13$, $p = .002$) see **Figure 5.4**.

> *R2: "To me, it's positive that it makes me more aware. You can't stick your head in the sand, the disease will catch up with you no matter what you do."*

In **Figure 5.3** is also presented that about one in two of our survey respondents (53%) state that they self-track because they enjoy it. However, they also think it is challenging to track; 58% find it difficult to know what to track and 61% find it difficult to know how to track. No more than 22% say that they try to track everything and 18% say they try to track all the time. In contrast, 61% say they track sometimes and that they track specific aspects of their PD. Persons with PD who have been diagnosed for more than five years are significantly more likely to try to track all the time (21%) than those who were diagnosed less than five years ago (13%) ($\chi 2(2) = 6.676$, $p = .04$), see **Figure 5.5**. In total 36% of persons with PD share their experiences of tracking with others, see **Figure 5.3**. Those who have been diagnosed for more than five years are significantly more likely to share with others (46%) compared to those diagnosed less than five years ago (23%) ($\chi 2(2) = 10.084$, $p = .006$), see **Figure 5.5**.

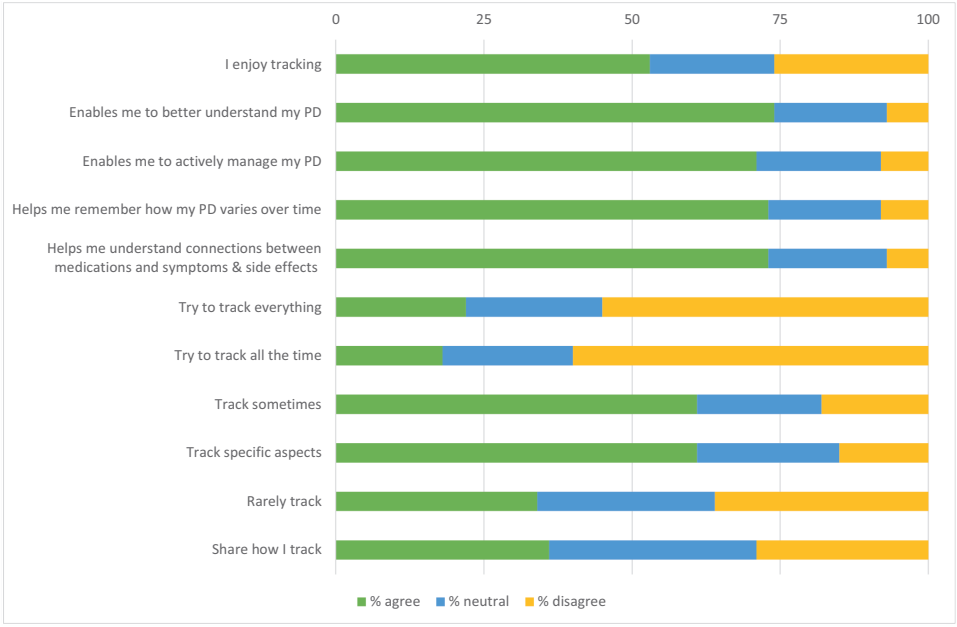

FIGURE 5.3: Why I self-track.

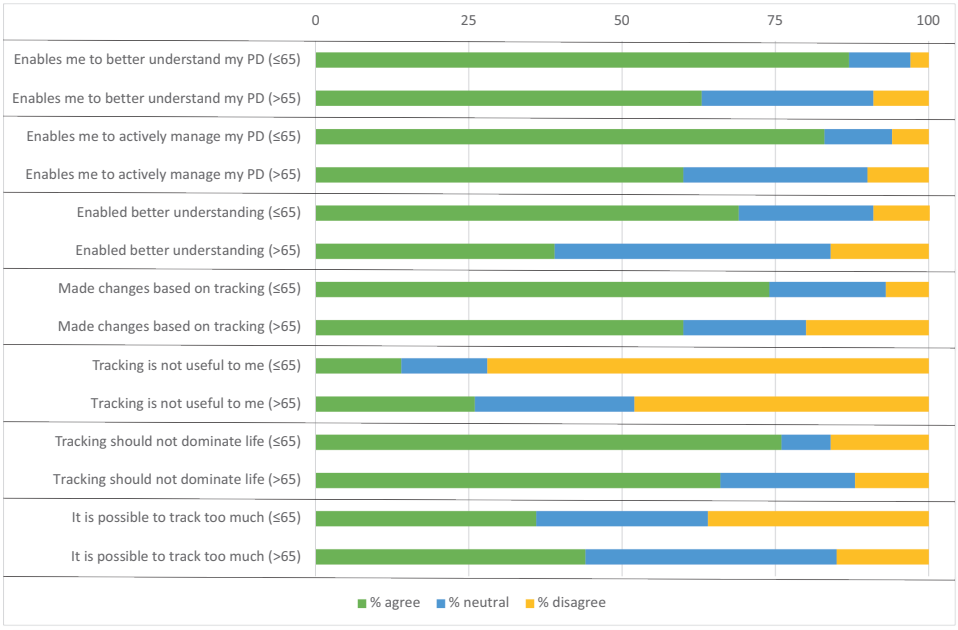

FIGURE 5.4: Significant differences between persons with PD younger and older than 65.

How and what I self-track

Our interview respondents described knowing what and how to track as important. The primary parameter to track among the participants was medication intakes, with the purpose to optimise timings. Other parameters important to measure were stress, diet and sleep. Different influencers were also acknowledged as a challenge in the interpretation of the collected data, and several participants described how important it was to understand these complexities in order to benefit from tracking.

> *R1: "It's important to take your medication right, at the right time. You can get a bad effect, it doesn't always mean that you need to increase your dose, it can mean that you need to make it more evenly distributed."*

The respondent with the most experience of tracking also expressed more abstract reasoning around how to capture the right measure when tracking, and what effects the choice of measurement can have. The same respondent also reflected on the challenges of defining more subjective measures to find a measure that is both efficient and reliable. What does it mean to me to "feel well"?

> *R1: "It was really hard work to constantly think about whether I didn't feel pain somewhere, if I didn't feel stiff and so on... It took over my life... So I realised*

that I have to register something else and I decided to make notes of when I am doing well instead, when my symptoms are on the level I want them to be."

Tools used for measuring include different kinds of activity trackers, smartphone apps for tracking sleep, exercise or similar. Several of the respondents used spreadsheet programs (e.g. Microsoft Excel) for gathering data and making graphs or other visual representations of the data.

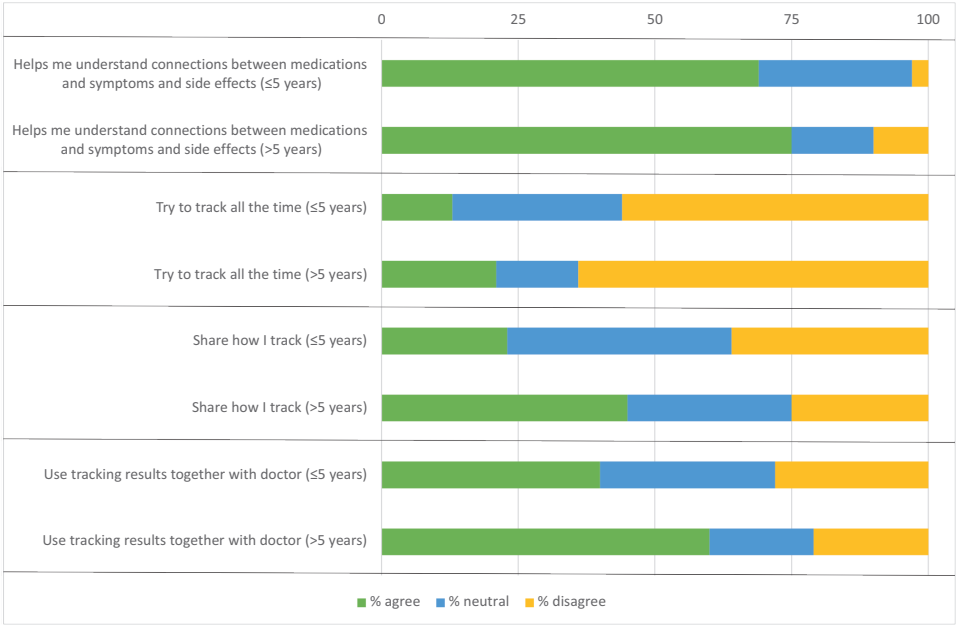

FIGURE 5.5: Significant differences between persons with PD diagnosed less or more than five years ago.

From the 180 included survey respondents, 49% had used some kind of technology, for their tracking. It could be a computer, smartphone, tablet, sensors or other devices, like smart watches, see **Table 5.3**. More frequently used was pen and paper (56%), and 74% had kept track in their head. Mode of tracking seems to be unrelated to gender, age, and education level. When it comes to time since diagnosis however, persons with PD diagnosed more than five years ago were significantly more likely to track using pen and paper (66%) than those diagnosed five years ago or less (41%) ($\chi 2(1) = 10.533$, $p = .001$), see **Table 5.3**.

The most common aspects to track in the survey were medication intake times (67%), medication types (62%), and physical activity/exercise (61%), see **Figure 5.6**.

TABLE 5.3: Mode of tracking.

I have experience of self-tracking using:	Technology		Pen and paper		My head	
	% agree	% disagree	% agree	% disagree	% agree	% disagree
Dx 5 years ago or less	44	56	41	59	75	25
Dx more than 5 years ago	53	47	66	34	74	26
Grand total	49	51	56	44	74	26

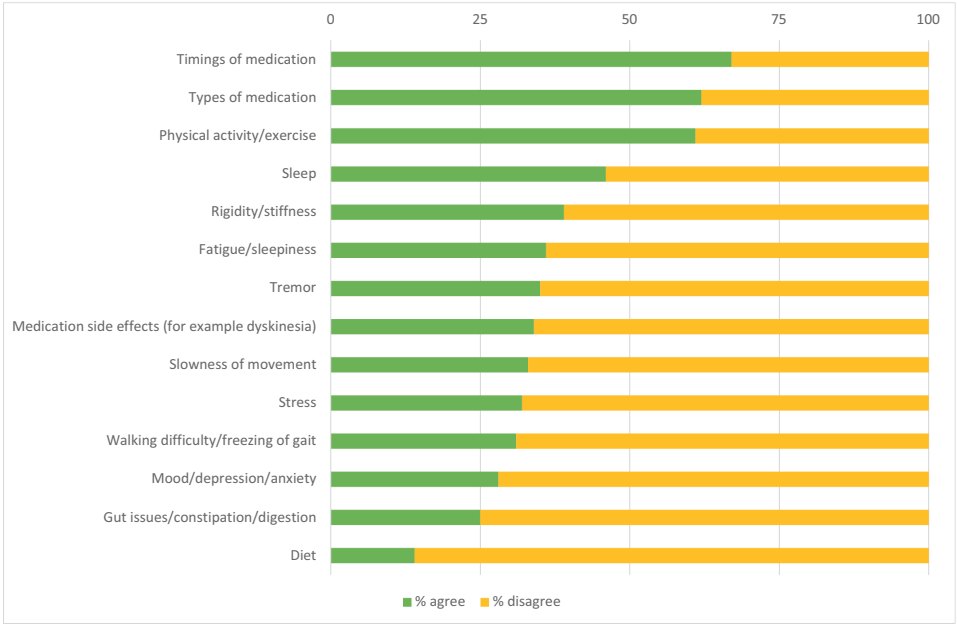

FIGURE 5.6: Aspects of PD tracked.

Lessons learned from self-tracking

Another category emerging from the data was lessons learned from self-tracking. The category includes insights gained from exploring tracking as well as barriers and challenges.

Several of the interview respondents mentioned that they had to learn a lot themselves to develop the understanding for the causalities and inferences necessary to really benefit from tracking.

R5: "I have gathered a lot of knowledge for myself in order to understand the connections. I found knowledge by reading, online, patient associations, conferences."

They had gained interesting insights and concrete results from their tracking and described how they had realised what was important for understanding their PD. For example how their medication was connected to sleeping patterns or a desire for sweets and how their ability to engage in physical activity varied over time. The respondents used the knowledge gained from tracking in different ways: for example for tweaking their medication regimen or for adjusting their food intake.

R6: "It's difficult to tweak medication timings, there are so many influencing factors; stress, food, lack of sleep, it's all inter-connected."

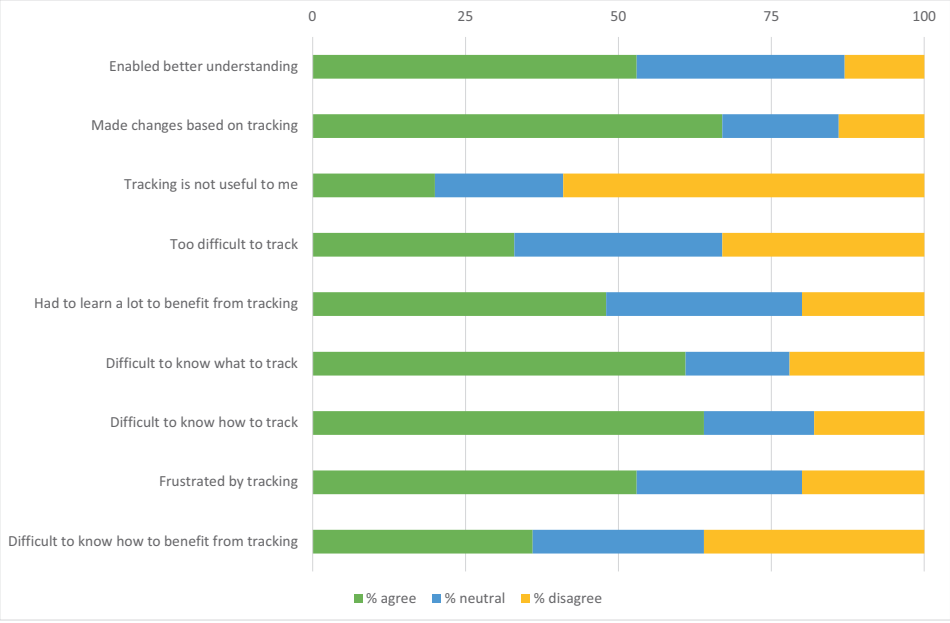

FIGURE 5.7: Lessons learned from self-tracking.

In the survey results, the ways the respondents utilise tracking varies: 51% said that they have made observations through their tracking that has enabled them to understand their PD better and 67%, said that they have made changes to their PD management as a result of tracking, see **Figure 5.7**. For persons with PD under 65, this rate was significantly higher (69 and 73% respectively) compared to those older than 65 (39 and

61% respectively) ($\chi 2(2) = 15.841$, $p = .0004$; $\chi 2(2) = 5.998$, $p = .05$), see **Figure 5.4**. One in two, 53%, were frustrated with how difficult it is to track and 36% think it is difficult to know how to make use of the results of tracking, see **Figure 5.7**.

Risks related to self-tracking

Potential risks related to tracking are the fixation of tracking and to let it take over your life. Most frequently mentioned by our interview respondents was the risk of persons with PD getting fixated or obsessed with tracking.

This can be related to effects of the disease itself and/or medication. The importance of finding a balance in life and not let the tracking get in the way of living was also mentioned. The respondents stressed that there has to be a balance between tracking to learn about your own condition and giving the disease too much focus.

> *R1: "I don't think you should be doing it all the time if you don't know what you want to use it for. Just tracking, that's pointless. You have to know why you're doing this."*

Our included survey respondents also see risks associated with tracking; one in two, 51%, think there is a risk of becoming obsessed with tracking PD, see **Figure 5.8**. A major part of respondents, 71%, do not want tracking to become too large a part of life (see **Figure 5.8**) and persons with PD younger than 65 are significantly more concerned (76%) than those older than 65 (66%) ($\chi 2(2) = 5.995$, $p = .05$), see **Figure 5.4**. In total 39% believe that it is possible to track too much (see **Figure 5.8**) and persons with PD older than 65 were more likely to agree with that statement (44%) than those younger than 65 (36%) ($\chi 2(2) = 10.677$, $p = .005$), see **Figure 5.4**.

On the issue of data privacy, our interview respondents in general expressed a trust in the healthcare system and did not see any major risks when it comes to sharing their data, neither with healthcare nor with other patients or caregivers. Sharing was considered positive as long as it was on their terms and with appropriate security measures in place. Our survey respondents were not reluctant to sharing data from their tracking; 72% were positive to sharing with healthcare and 61% were open to sharing with anyone who is interested, see **Figure 5.8**.

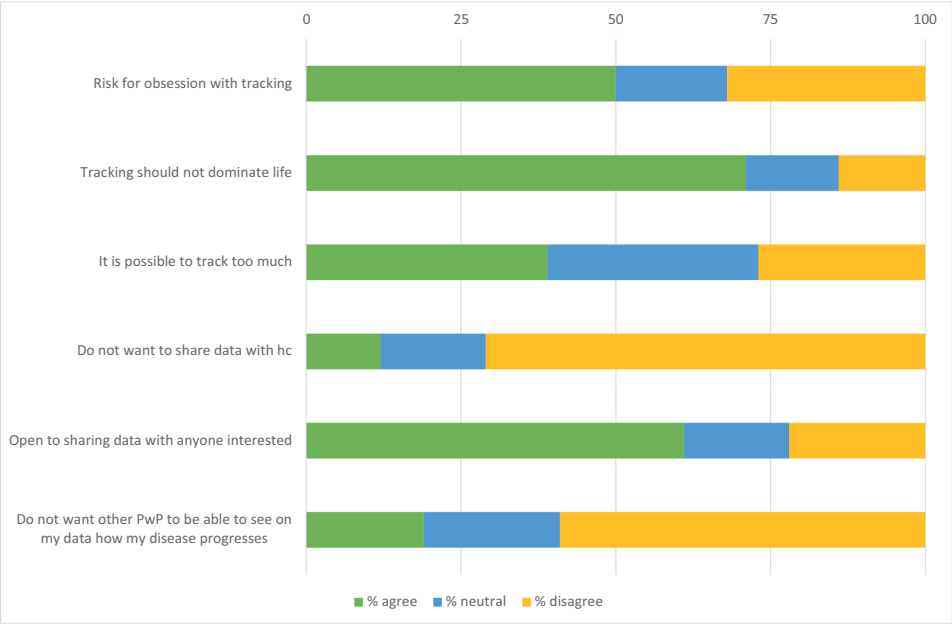

FIGURE 5.8: Risks related to self-tracking.

5

Self-tracking and healthcare

The practise of tracking can influence the relationships with healthcare, both in a positive and a negative way and this is described in the category *self-tracking and healthcare*.

Some of our interview respondents used tracking results as a trigger to contact healthcare to handle worsening symptoms. They expected healthcare to be interested in tracking data, since this kind of information is difficult for healthcare to come by without patients collecting the data themselves. One expected benefit was that it would save time at the doctor's visit if persons with PD collected data beforehand. Tracking as memory support was considered important in preparation for clinical encounters and to give their clinicians an accurate account of their disease status since the previous appointment.

> *R7: "Sometimes I have been allowed to present my tracking but there doesn't seem to be much interest from healthcare. I think it has to do with the attitudes of doctors, I get the feeling that they want to do their assessments without involving my tracking."*

In total 36% of our 180 included survey respondents used tracking to decide if they needed to visit their physician and 53% said that they track to prepare for healthcare visits, see Fig. 9.

Women are significantly more likely to track to prepare for healthcare visits than men (63% compared to 43%) ($\chi 2(2) = 8.588$, $p = .02$), see **Figure 5.10**.

The interview respondents expressed a wish to collaborate with healthcare around tracking. There can be a potential benefit from making use of data that are collected in-between healthcare encounters. At the same time, our respondents showed understanding for the challenges that healthcare professionals face, which can explain healthcare professionals' hesitation for encouraging tracking. In general, the respondents appreciated that it can be difficult for healthcare professionals to change the way they are working. Financial concerns were also mentioned, as well as the need for bringing context to the tracking data. It was also expressed that both healthcare and persons with PD need better tools and support for analysing tracking data. Several of our respondents demonstrated how the tracking they undertook and the knowledge they gained from this enables them to be more active in communicating with healthcare.

> *R4: "... and the doctor says...: 'Let's replace [medication X] with [medication Y], and you can take it in the evening'. 'But if I do that, I won't be able to work because I will have more tremor in the mornings.' 'Yes' she said. 'But, can't I take [medication X] in the morning instead?' I said. 'And I will be able to work', I said. And if I hadn't known this, I would have taken her suggestion and I would have functioned less well in daytime, and maybe I wouldn't have been able to work as much."*

Figure 5.9 shows that more than one in two persons with PD in the survey (53%) showed their tracking results to their physician. However, only 32% said that their physician encourages them to track and only 21% said that their physicians are interested in their tracking. In total 52% of persons with PD said that their tracking results are used in the clinical encounter to make treatment decisions and for those diagnosed more than five years ago, the rate was significantly higher (60%) compared to those diagnosed less than five years ago (40%) ($\chi 2(2) = 7.299$, $p = .03$), see **Figure 5.5**. As many as 82% of our respondents thought that healthcare needs to find better ways to assess PD on an individual level, see **Figure 5.9**.

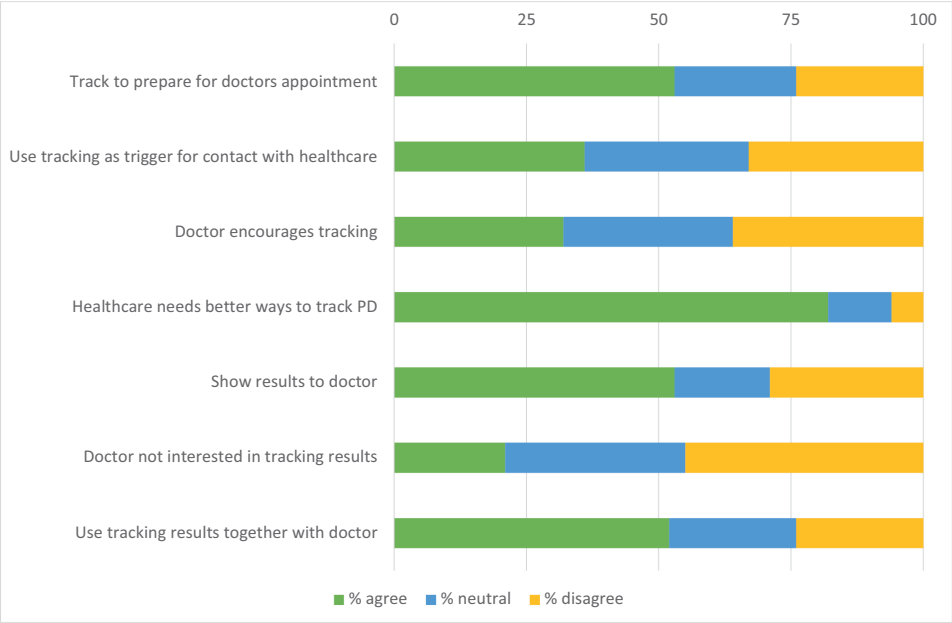

FIGURE 5.9: Self-tracking and healthcare.

Model for self-tracking in PD

Figure 5.11 shows the proposed model based on the analysis of our study results. Core concepts in the model relate to the motivation and drive to self-track (Why I self-track), skills and tools required (How and what I self-track), and the knowledge produced from self-tracking (Lessons learned from self-tracking). Risks were mentioned across all these areas, as were the mixed experiences of utilising self-tracking in collaboration and communication with healthcare.

When working with the categories, we found that they were connected via a theme, going across the data. The respondents' descriptions and experiences demonstrated that there is a lot of work associated with self-tracking and that the respondents expressed both benefits and burdens. We found that an overarching theme emerged, namely Balancing benefits and burdens. Examples of mentioned benefits and burdens are given in **Figure 5.11** and some of the strategies for finding this balance that emerged from the interviews will be addressed in the Discussion section.

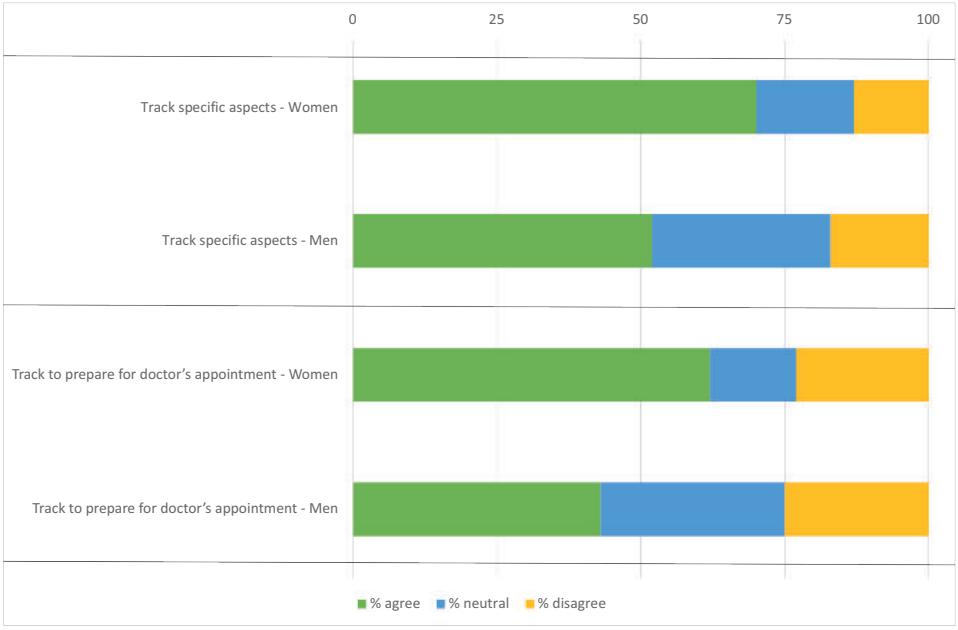

FIGURE 5.10: Significant differences between women and men.

DISCUSSION

The aim of this study was to explore the experiences and opinions of people with PD in Sweden on self-tracking and the results presented above indicate that it can be both rewarding and challenging. We found that the "average PD self-tracker" track different aspects of their disease and treatment, for example timings and/or types of medication, and exercise or physical activity. Mostly they track only in their head, but sometimes they also make notes on paper or use technology. They see tracking as a tool they can use to better understand and actively manage their PD. They track specific aspects occasionally, for example to help them understand how medications relate to symptom relief and side effects and to remember how their disease varies over time. Tracking can be frustrating, it can be difficult to know what and how to track and they strongly believe that healthcare needs better ways to assess PD. They track to prepare for doctor's appointments and use the tracking results together with their doctor. When it comes to sharing their tracking data they are open to doing so with healthcare and other people interested. They do see a risk for becoming obsessed with tracking and think it is important that tracking does not dominate life.

We propose that self-tracking as described by our respondents could be seen as a personal improvement project, aiming to understand and improve one's own health

and disease management. Continuous quality improvement in healthcare is often based on the model for improvement, the PDSA cycle, introduced by Edwards Deming and further developed by Nolan et al. [26]. The PDSA cycle approach is an iterative process for learning and improving complex systems and resembles the process of an individual learning about their own health in order to improve it. The approach has in fact also been used in trying to improve individual health in chronic disease [27]. Three fundamental questions form the basis for improvement, according to [26], namely:

1. What am I trying to accomplish?
2. How will I know that a change is an improvement?
3. What changes can I make that will result in improvement?

The PDSA approach resonates well with the categories emerging from our study results, see **Figure 5.3**. We posit that our category "why I self-track" corresponds to the first of the fundamental questions, our category "how and what I self-track" to the second question, and our category "lessons learned from self-tracking" to the third.
The theme of balancing benefits and burdens that we found in our data is not as clearly expressed in the PDSA cycle. It is however present in the notion that it has to be possible to determine whether an improvement has occurred – otherwise the change is potentially only a waste of efforts and resources. When patients engage in personal improvement related to their health the stakes are even higher and require other strategies for finding the balance between benefits and burdens, as we will discuss in more detail below.

Women in our survey are more likely than men to track specific aspects as well as to track in preparation for doctors' appointments. For persons with PD younger than 65, there are some significant differences compared to the group as a whole. When it comes to making use of tracking, people in the younger group are more likely to have made changes to for example their treatment as a result of tracking. Younger persons with PDs are also more likely to disagree with the statement that tracking is not useful for them. Our results also indicate that the older group are more likely to agree that it is possible to track too much. Time since diagnosis seems to be the most important factor when it comes to attitude and use of self-tracking. In total, 64% of our survey respondents have experience from tracking. The characteristics of trackers and non-trackers are not significantly different when it comes to age, gender distribution, or education level. The groups are however different when it comes to time since diagnosis; persons with PD diagnosed more than five years ago are significantly more likely to self-track than persons diagnosed for five years or less. Furthermore, persons with PD diagnosed more than five years ago are significantly more likely to consider tracking helpful, to share their learnings from tracking with others, and to use tracking together with their doctor, than persons diagnosed a shorter time.

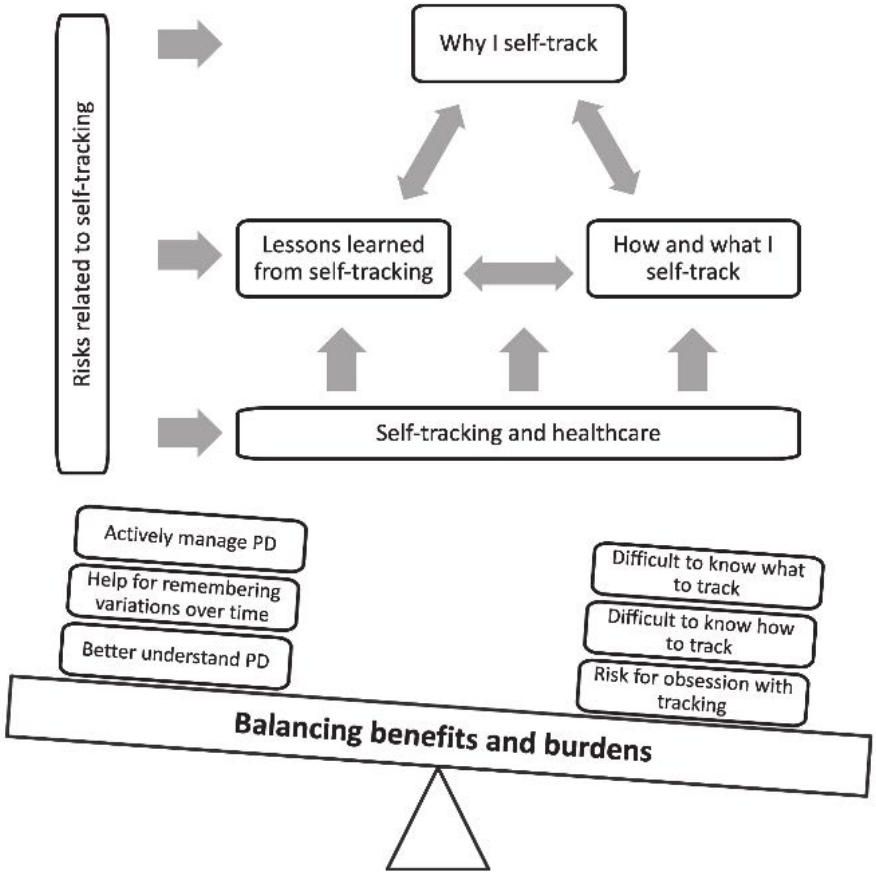

FIGURE 5.11 Balancing benefits and burdens.

Research on tracking in other conditions and contexts has made findings of relevance to our study. A study exploring tracking in multiple chronic conditions found that patients considered tracking to be "illness work" and that it continuously reminded them of their conditions [28]. This goes in line with our findings and it is possible that the insight from one of our respondents of tracking well-being rather than problems could make the work less burdensome. It could be one important strategy for finding the balance by reducing the experienced burden that focus on the illness creates. Sharon [29] has done work in self-tracking and found that there seems to be important considerations to make concerning autonomy, solidarity, and authenticity in relation to self-tracking as an element in personalised healthcare.

Persons with PD in Sweden make the same conclusion as U.S. adults with other chronic conditions; that tracking affects the way they manage their health and communicate

with healthcare providers, according to the knowledge they gain from tracking [30]. In a healthcare context, PD is often seen as mainly affecting movement whereas persons with PD often state that non-motor symptoms are at least equally bothersome. However, self-tracking and the right tools for analysis could help also healthcare to see PD in a wider perspective. A previous study of perceptions of the use of technology for PD and epilepsy indicated that both healthcare professionals and persons with PD saw potential benefits for better understanding of the disease and improved disease management [14]. In that study, as in ours, persons with PD saw potential for improved selfcare, including medication management. Healthcare professionals in the previous study however saw risks with persons with PD adjusting their medications themselves [14]. Our study did not elicit the views of healthcare professionals but our respondents in both the interviews and the survey clearly expressed a desire to collaborate with healthcare and a frustration at the lack of effective tools. A large majority (82%) of our survey respondents expressed that healthcare needs to find better ways to assess PD on an individual level, stressing the importance of utilising tools such as self-tracking to achieve personalised healthcare. By ensuring that self-tracking is integrated in and accepted by healthcare the benefits of self-tracking could increase contributing to improving clinical management based on selfcare by persons with PD. The findings of our study support the views of previous studies [28,29] that the actual user perspective needs to be more in focus for self-tracking to truly support the much needed transformation of healthcare.

Limitations

Our study is not without limitations. Our focus was on persons with PD in Sweden and the generalisability of our results is unclear. Seven interview respondents can be considered a small number. We were however less interested in representativity of all persons with PD and more interested in exploring the views and opinions of persons of PD with experience of self-tracking. We did reach saturation in the context of self-tracking, which indicates that the numbers were sufficient for the purpose of this mixed-method study. Another limitation concerns the survey. We chose to collect responses online which is a method generally suited for people who are already active and engaged. Furthermore, our PD trackers survey respondents are relatively well educated, 62% have studied for more than 12 years. This can be compared to the general level of the Swedish population, which is 30–35% [31]. Above factors could mean that our results show more of what persons with PD who are already interested in self-tracking think and underestimates the problems involved. Also, since one of the researchers (SR) has PD herself and has been conducting self-tracking for a long period of time, there could be a bias in underestimating the challenging aspects. We found however that risks and challenges were something that comes with long experience of tracking.

Future research

Our study suggests that there is potentially a lot to be learned from persons with PD self-tracking on their own initiative and that the tools needed at least partly have distinctly different characteristics from tools used by and in healthcare. In this field, we have identified possible future work in the design and implementation of tools for measuring the "right" thing as well as for storing, analysing, visualising, and sharing data. We have also identified a number of other strategies that self-tracking patients apply to reduce the burden of tracking, e.g. focusing on tracking positive aspects rather than negative, or clearly limiting their tracking in both time and focus. It would be of interest to further explore how widely spread these strategies are and how effective they are in reducing the burden of self-tracking. We believe that the PDSA methodology could be a useful tool in exploring these issues further.

Another topic for further research is looking into the group that do not track. What can we learn from them? What are their reasons for not tracking?

We have also identified a neglected area in education related to self-tracking, both for persons with PD and healthcare professionals. With a better understanding of the needs for knowledge, both theoretical and practical, the benefits of self-tracking can be realised in a better way. Future work in this area includes for example identifying appropriate methods and actors for education as well as organisational and funding issues.

Data from self-tracking efforts by individuals can also potentially be used for systematically improving healthcare and research, ultimately enabling personalised medicine. This would lead to a clearer focus on secondary prevention, which has the potential of improving health. This potential warrant further studies relating to, for example, how self-tracking could influence health economical aspects, both in healthcare as well as within the society.

CONCLUSIONS

To the best of our knowledge, this is the first study exploring why and how persons with PD self-track and despite the limitations mentioned above, we believe that our results are an important contribution to extending the knowledge in the field of self-tracking in PD.

The extremely individual nature of PD makes it highly suited for self-tracking efforts and persons with PD who have tried think it entails both important benefits and burdens. The main identified benefits are that self-tracking can lead to a better understanding

for persons with PD of their own specific manifestations of PD and contributes to a more effective decision making regarding their own selfcare. The process of self-tracking also enables persons with PD to be more active in their communication with healthcare. This is important, especially considering the limited time they have with healthcare. We believe that this study's main contribution is the insight that tracking takes a lot of work and there is a need to find the right balance between burdens and benefits. This balance can be as individual as the symptoms of PD itself, yet some strategies have been identified in this study; to focus on positive aspects rather than negative, to limit the focus of self-tracking both in time and scope, and to increase the benefits through improved tools and increased use of self-tracking results in the dialogue with healthcare. However, we still need a clearer understanding of these burdens and benefits, from the individual perspectives of every stakeholder, mainly persons with PD, healthcare professionals, and researchers respectively.

Ethics approval and consent to participate

Based on an application, the Regional Ethical Review Board in Stockholm ruled that the legislation regarding ethical review is not applicable to this study (according to decision 2015/1572–31/4). However, all participants were informed about the study and how their personal data would be handled anonymously, published and stored. All interview participants signed a consent form for their participation in the study. The survey participants were informed that responding to the survey implied consent to participate

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APPENDIX 5.1

Interview guide self-tracking

Information about the study

a. OK to record?
b. Time: no more than 1 hour
c. Review the form for information and consent. Point out that participating in the interview is voluntary, they are free to not respond and free to interrupt the interview at any time.

Background information:

1. "Can you tell me a bit about yourself?"
    a. Age? Family situation? Where do they live? Profession?
    b. Disease history? Time of diagnosis? The situation around the diagnosis?
    c. Main symptoms? Which are most troublesome?
2. "Can you tell me about the healthcare you receive for your Parkinson's disease?
    a. Frequency? Which aspects are you pleased with? Less pleased with?
    b. What does your current treatment consist of?
        i. Medications?
        ii. Other treatments?
    3. "Can you tell me about something that you do to improve your wellness that is not prescribed by healthcare?"

Self-tracking: Tell them what we mean by self-tracking, that we mean both things you measure using apps, devices and similar, but also observations made using pen and paper and even things you "just" track in your head.

1. Self-tracking:
    a. "Can you tell me what you know about self-tracking?"
    b. "Have you tried it yourself?/Are you interested in trying?"
    c. "Why are you interested/not interested?"
    d. "Which expectation/concerns do you have?"
2. Self-tracking and healthcare:
    a. "Do you have any thoughts around self-tracking and healthcare?" If they cant think of anything, nudge them by saying for example: "Do you think that healthcare would be interested in self-tracking?"
    b. "What are your thoughts around sharing self-tracking data with others, for example healthcare, other patients, family members?"
    c. "Do you see any risks/benefits/downsides?"
    d. "Can you describe what you would like the collaboration between you and healthcare around your Parkinson's disease to look like?"
3. "Is there anything you would like to add?"

APPENDIX 5.2

Survey questions

A1. How long ago were you diagnosed with PD?

- Within the last year
- 1-5 years ago
- 6-10 years ago
- 11-15 years ago
- 16-20 years ago
- More than 20 years ago

A2. Are you... Male Female Prefer not to say

A3. Your age?

- Younger than 25
- 26-35 years
- 36-45 years
- 46-55 years
- 56-65 years
- 66-75 years
- 76-85 years
- 86 years or older

A4. Highest completed education?

- Compulsory school (<9 years)
- Upper secondary school (9-12 years)
- University (>12 years)

A5. County council of residence

B1. I use (or have in the past) the following methods to keep track of aspects relating to PD. (tick all that apply)

- Computer, smartphone (incl apps), tablet etc
- Sensor based technology, for example activity tracker, smartwatch etc
- Pen and paper
- I track in my head
- Other methods (please comment below)

B2. I would like to keep track of the following aspects relating to PD. (tick all that apply)

- Tremor
- Slowness of movement

- Rigidity/stiffness
- Walking difficulty/freezing of gait
- Mood/depression/anxiety
- Fatigue/sleepiness
- Gut issues/constipation/digestion
- Types of medication
- Timings of medication
- Medication side effects (for example dyskinesia)
- Diet
- Exercise
- Sleep
- Stress
- Other aspects of PD (please comment below)

B3. I keep track of (or have in the past) the following aspects relating to PD.

- Tremor
- Slowness of movement
- Rigidity/stiffness
- Walking difficulty/freezing of gait
- Mood/depression/anxiety
- Fatigue/sleepiness
- Gut issues/constipation/digestion
- Types of medication
- Timings of medication
- Medication side effects (for example dyskinesia)
- Diet
- Exercise
- Sleep
- Stress
- Other aspects of PD (please comment below)

B4. I keep track (or have in the past) of aspects relating to PD for the following reasons. Please indicate whether you agree or disagree with the following statements. (Options are: Strongly disagree, Disagree somewhat, Neither agree or disagree, Agree somewhat, Strongly agree)

- I enjoy tracking
- Tracking enables me to understand my PD better
- Tracking enables me to take an active approach in the management of my PD
- Tracking enables me to remember how my PD fluctuates over time
- I use tracking to prepare for healthcare visits

- I don't think tracking is useful for me
- Tracking helps me understand how my PD medications influence my symptoms and side effects
- Other reasons (please comment below)

B5. My approach to tracking of aspects relating to PD is (or has been in the past). Please indicate whether you agree or disagree with the following statements. (Options are: Strongly disagree, Disagree somewhat, Neither agree or disagree, Agree somewhat, Strongly agree)

- I try to track everything
- I try to track all the time
- I track sometimes
- I track specific things
- I think it is too difficult to track
- I rarely track my health
- I have had to learn a lot about PD to be able to benefit from tracking
- I find it difficult to know what to track
- I find it difficult to know how to track

B6. My use of tracking of aspects relating to PD is (or has been in the past). Please indicate whether you agree or disagree with the following statements. (Options are: Strongly disagree, Disagree somewhat, Neither agree or disagree, Agree somewhat, Strongly agree)

- I have made observations based on my tracking that have helped my understand me PD better
- I have changed things (for example medication type and/or timings, diet, exercise regimen etc) as a result of tracking
- I use tracking to decide if I need to contact healthcare professionals
- At some point in the past, I have been frustrated by how difficult it is to track different aspects of my health
- I share how I track my health with other people.
- I find it difficult to understand how to make sense of the things I track

B7. Tracking and healthcare. Please indicate whether you agree or disagree with the following statements. (Options are: Strongly disagree, Disagree somewhat, Neither agree or disagree, Agree somewhat, Strongly agree)

- I show my physician results from my tracking
- My physician is not interested in the results of my tracking
- My physician and I use results from my tracking in our discussions about my treatment

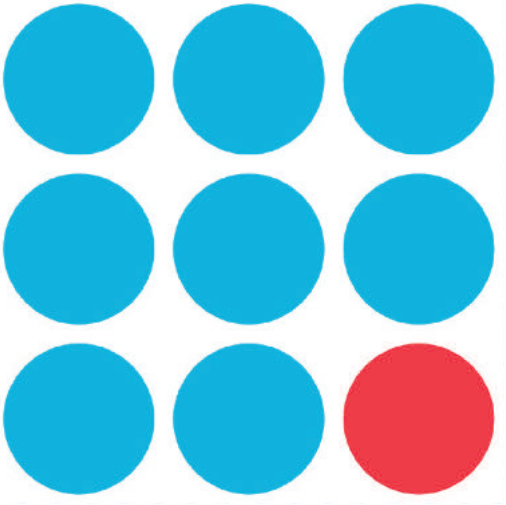
- My physician encourages my tracking
- I think healthcare should find better ways to evaluate/assess PD on an individual level

B8. Risks and challenges with tracking. Please indicate whether you agree or disagree with the following statements. (Options are: Strongly disagree, Disagree somewhat, Neither agree or disagree, Agree somewhat, Strongly agree)

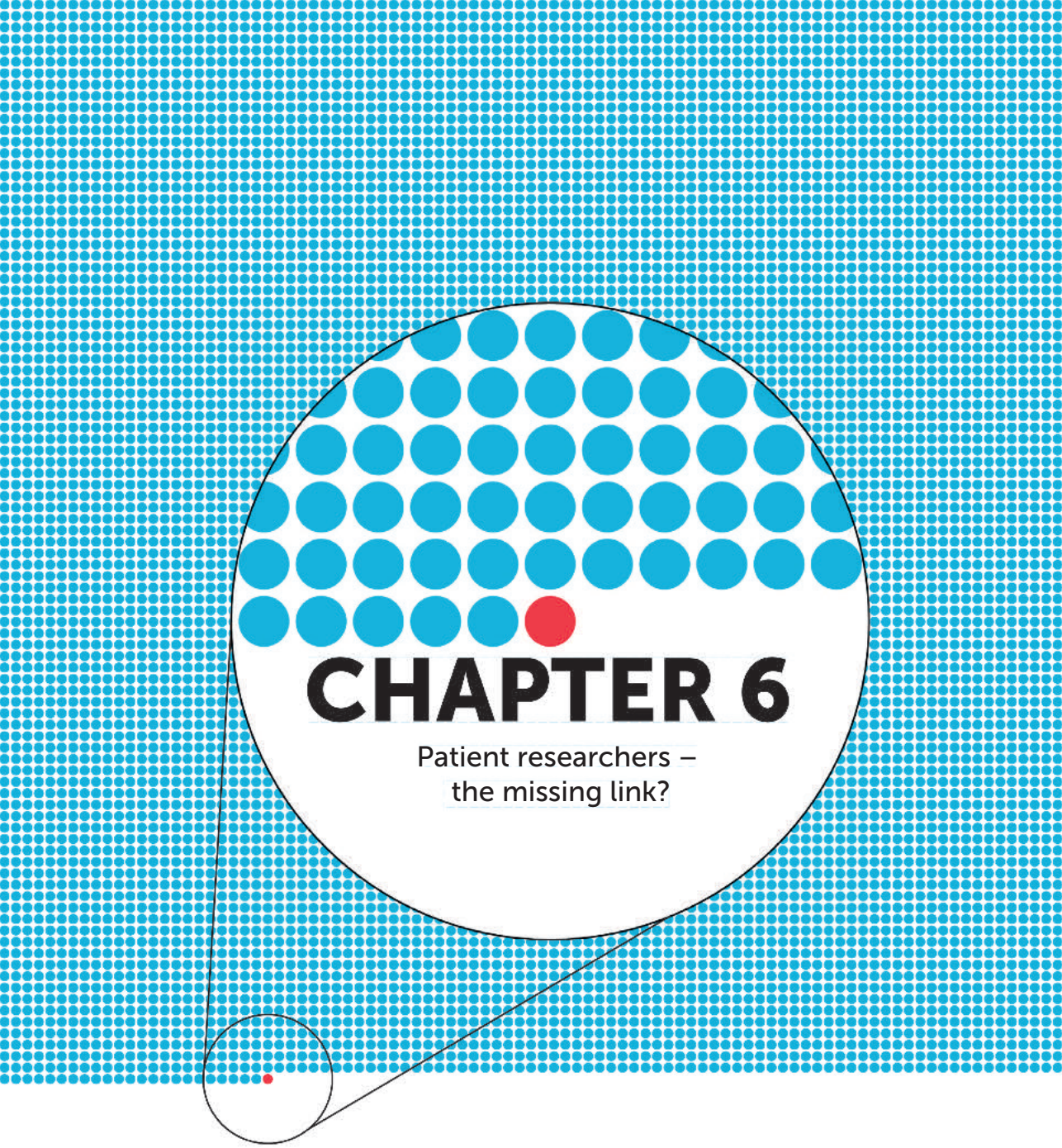
- I think there is a risk for becoming obsessed with tracking PD
- I don't want tracking to get in the way of living
- I don't want to share my tracking data with healthcare
- I am open to sharing all my tracking data with anyone interested
- I don't want other people with PD to be able to see data of how my PD is progressing
- I think that you can track too much

PART III

PATIENT-LED RESEARCH



My least favourite symptom of PD is freezing-of-gait, or FOG for short. It is a strange type of gait disturbance that is one of the many symptoms of PD. As my disease has progressed over time, my gait has become increasingly affected. What does FOG look like? Well, before I had experienced it myself, I couldn't understand why some persons with PD would sometimes suddenly stop walking and just stand still, looking literally as if they were frozen to the ground or glued to the floor. Having experienced it myself, I now know more about the situations and contexts where my risk of "freezing" is higher, but it is still a bit of a mystery. During a FOG episode, my steps become increasingly quicker and shorter until I stop, then appear frozen to the ground. Sometimes the momentum of my upper body makes regaining control of balance impossible because my centre of gravity is already too far in front of the toes of my feet, and I will fall forward. I say fall, but it is much less dramatic, more like kneeling on my right knee. In a way, I think that FOG is my brain's emergency brake. It can occur when I'm walking and too much happens at the same time, or when my stress level is elevated. For example, when I'm hurrying to cross a road before the light changes, or when something unexpected happens, like a sudden movement or a sudden sound. In such cases, my brain sometimes decides to try to protect the body from what it interprets as being something potentially dangerous. It does this by stopping the body from moving forward. When FOG happens, my mind goes completely blank, it feels like my brain is short circuited.



CHAPTER 6

Patient researchers – the missing link?

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PATIENT RESEARCHERS - THE MISSING LINK?

I have over 35 years of experience of living with a complicated chronic disease. As early as 13 years of age, I noticed that my body did not always respond to the signals from my brain or move in the way I wanted it to. Almost two decades later, I was diagnosed with Parkinson's disease.

In 2010 I decided that I wanted to combine my training in engineering with my patient experiences to improve the situation for myself and others with chronic diseases. I have made the management of my own disease my topic of research, conducting a doctoral degree in self-tracking and personal science for Parkinson's disease at Radboudumc in The Netherlands and Uppsala University in Sweden. I am a 'patient researcher'.

Amazing progress has come from the traditional model for biomedical research. Since the first reported clinical trial in 1747, we now have access to vaccines and effective treatments for many diseases that used to be deadly. But in all that glorious history of research, patients' contributions have been seen mostly as only passive; patients have been considered only as the subjects of studies that are designed and conducted by clinical researchers, who almost always lacked lived experience of the disease they were studying.

Today, it is well known that patient partnerships improve research. An increasing body of evidence demonstrates that the priorities of patients, across diseases, are often radically different from what clinicians think that they are, and that clinicians are often not addressing the issues that really matter to patients. A study estimating the financial value of patient engagement in the context of oncology found that an investment of US$100,000 in patient engagement can yield a gain of several hundred times as a result of improved patient experience [1]. There are many organizations around the world working to develop and promote patient partnerships in research and healthcare improvement, including The Patient-Centered Outcomes Research Institute (USA) and The James Lind Alliance (UK).

But what would happen if we took this one step further? What might happen if more patients join me to become 'patient researchers'?

Patient researchers are people with lived experience of a disease, disability or other health challenge who are openly using those experiences in conducting research within academia or in other contexts. A patient researcher can also be a family member, partner or similar of the person living with the disease.

There is great potential for patient researchers in the improvement of biomedical research. I would not be able to manage my Parkinson's disease as well as I do without all the knowledge I have acquired through my patient-researcher efforts, and people often tell me that my work has also helped others, both patients and researchers.

Being a patient and a researcher means having a foot in each camp. I have a deep knowledge of the burdens of living with a life-altering condition and at the same time I have an understanding of the ins and outs of the academic system. This means that I can use my dual roles and translate between the two communities. I take my role seriously and am careful to honor the trust that my patient peers have in me. Of course, this duality also comes with challenges: it is sometimes difficult to balance the needs of the community with the needs of the academic system. There are of course researchers without lived experience who actively engage with patients and do a great job. Even so, one benefit of being a researcher openly using my lived experience of chronic disease is that I can often connect quickly with other patients, across diagnoses, in a joint understanding of the limitations that come with our individual diseases

But patient researchers are receiving a lot of pushback in the academic world. Our contributions are questioned as being unrepresentative, invalid and unobjective. Some of these objections are definitely worth exploring further. But some should rightfully be chalked up to the rigidity of the conservative biomedical system. I have, for example, been told: "But Sara, why would patients want to do research on themselves anyway? Wouldn't it be better if you all just give your data to a proper researcher?" And in a way, I can understand that comment: in conventional research, patients are just subjects in the study; they are not researchers. But in my opinion, the system needs to change.
The positive contributions of patient researchers should be formally recognized, patients should be encouraged and supported to take the lead of research projects, and colleagues without lived experience should be educated to appreciate the contributions of their patient-researcher colleagues. There has been progress in that direction. For instance, the University of Oxford recently awarded the first two scholarships for patient-led research [2]. However, there is a long road ahead. I hope that the growing community of patient researchers can get the credit and support we need to continue developing.

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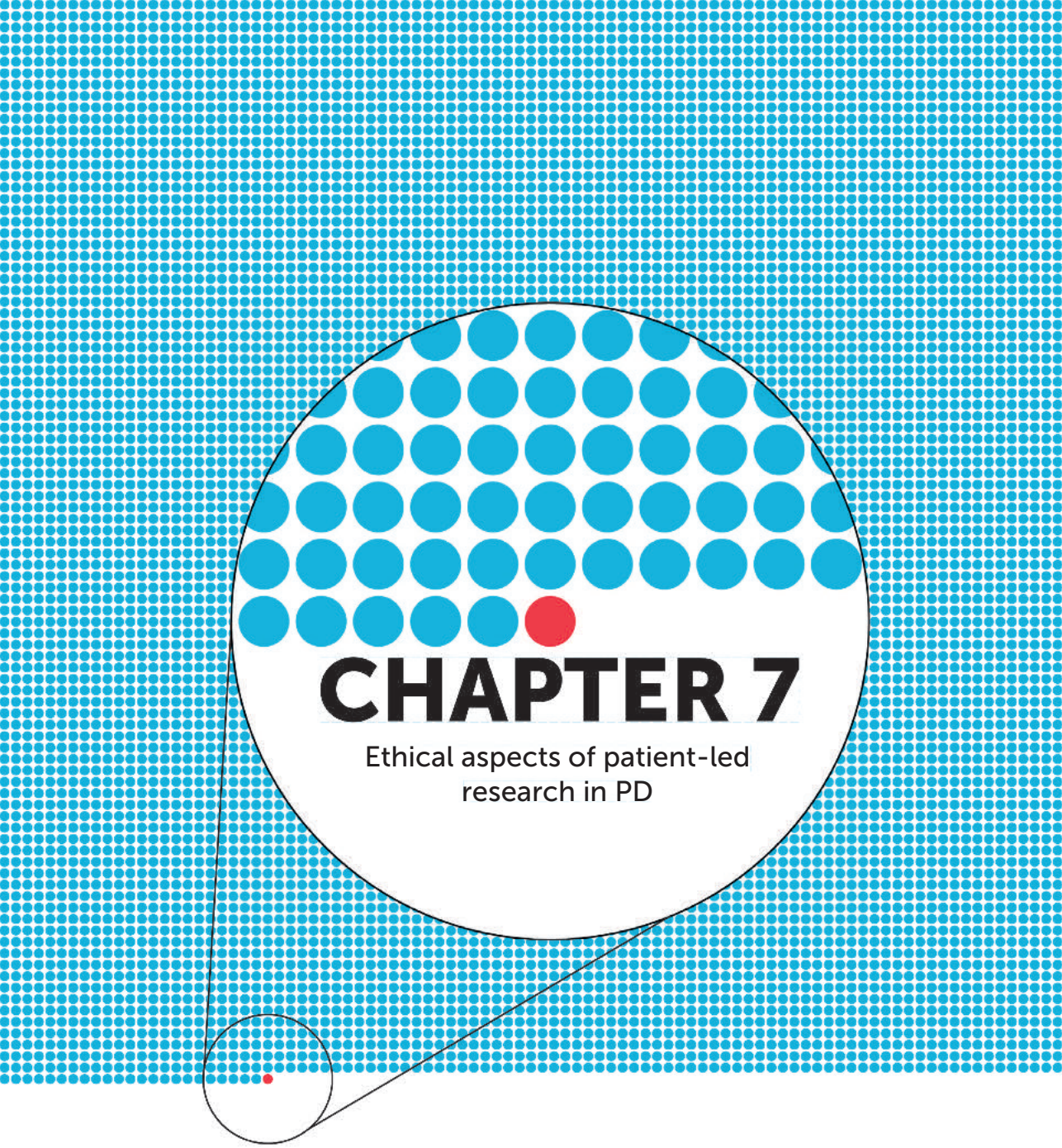

CHAPTER 7

Ethical aspects of patient-led research in PD

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ABSTRACT

Using Parkinson's disease as an exemplary chronic condition, this Commentary discusses ethical aspects of using self-tracking for personal science, as compared to using self-tracking in the context of conducting clinical research on groups of study participants. Conventional group-based clinical research aims to find generalisable answers to clinical or public health questions. The aim of personal science is different: to find meaningful answers that matter first and foremost to an individual with a particular health challenge. In the case of personal science, the researcher and the participant are one and the same, which means that specific ethical issues may arise, such as the need to protect the participant against self-harm. To allow patient-led research in the form of personal science in the Parkinson field to evolve further, the development of a specific ethical framework for self-tracking for personal science is needed.

INTRODUCTION

Parkinson's disease (PD) is a complex neurodegenerative condition displaying a wide range of motor and non-motor symptoms that are generally challenging to manage using available medical interventions [1]. This recognition has further stimulated the important ongoing development towards greater selfcare and patient participation in healthcare [2,3]. Indeed, persons with PD have to manage their condition and treatments on their own for most of the time. Examples include the need to ascertain that medically prescribed interventions are followed adequately, but also the responsibility to implement lifestyle interventions, such as exercise and a healthy diet. Additionally, there is an increasing emphasis on self-tracking, as an important way of detecting relevant disease complications in a timelier manner, also to monitor patients more closely in their own home living environment. However, persons with PD are mostly left in the dark during this process of selfcare, having to operate largely without suitable tools provided by the healthcare system, and without generally accepted biomarkers that could be monitored to inform their decisions. Consequently, active, engaged, and knowledgeable patients now increasingly take matters into their own hands by using self-tracking to perform research on themselves. Sometimes, these activities challenge the current system for ethical oversight and approval of research [4].

Frameworks for ethical conduct in clinical research have evolved over time, as is apparent from the use of the term "subjects" in an article published in 2000 [5], instead of, as is more common today, "participants". But what happens when individuals move beyond actively participating in clinical research to using empirical methods to improve their own selfcare? What happens when people managing health challenges on a daily basis, also known as patients, make use of the possibilities of the Internet and other technological developments to conduct their own research? To what extent do current ethical frameworks apply to these upcoming practices? Do specific ethical challenges emerge when individuals also intend to disseminate their findings by publishing them in a scientific journal?

Here, we will briefly present the emerging field of personal science and examine some of the main ethical considerations related to the use of self-tracking in personal science as well as in clinical group research. The practice of personal science has similarities to for example the fields of mHealth and citizen science but, as we will demonstrate, also evokes specific ethical challenges. For the discussions in this paper, we will focus on PD as an exemplary chronic condition, but the perspectives offered are likely to also be relevant for the ethical challenges of personal science for a wide range of other chronic diseases.

Personal science

Recently the concept of *personal science* as a framework for research has been introduced. Personal science has been described as: *"the practice of using empirical methods to explore personal questions"* [6], *"self-directed N-of-1 studies"* [7], *"an interest in collecting data about their own bodies or lives in order to obtain insights into their everyday health or performance"* [8]. Based on these key references, personal science is here defined as *the practice of exploring personally consequential questions by conducting self-directed N-of-1 studies using a structured empirical approach*. This practice is utilised by people with different backgrounds and health statuses, and also applied by people who are confronted with challenges and limitations as a result of chronic and progressive diseases, such as PD.

A key method for collecting data in personal science is self-tracking: "a process of deliberately collecting and structuring observations about one's own life" [6]. The phenomenon is as old as humankind and has emerged broadly and evolved along with the unfolding developments in technology and digitalisation. Its societal impact has been shown in the context of what has been referred to as the Quantified Self movement [6]. The wide availability of sensors, wearable devices and smartphones enables data to be collected about most aspects of our lives, including our health [9]. Of note, although self-tracking can be aided by technology, it can also be done simply using pen and paper [9].

Personal science can include both observational and interventional study designs. The generalisability of the approach can vary; the specific methods used by a single participant (namely measurements, data collection, evaluation, etc.) can potentially be generalisable to other persons dealing with similar health issues. For example, a custom-made app which can successfully track tremor in one particular person with PD can likely be extended to other persons with PD as well. In contrast, interventions that have a demonstrable effect for one individual still require very careful considerations before applying them to someone else.

Personal science can be practiced at different levels of impact. At the first level, the practice is intended to address issues identified by a given individual and to inform and improve the process of selfcare for just this person. Many of us already perform this type of investigations, for example when using commercially available activity trackers as a tool to be informed about and sometimes even improve physical activity. For personal science of that kind, ethical considerations are largely straightforward, and it will therefore not be the main focus of this paper. In contrast, it is in particular when experiences from personal science projects are publicly disseminated, for example in lay language on social media or in scientific publications, and thereby can lead to other

people being influenced, that specific ethical challenges emerge. Personal science that is publicly disseminated has similarities with citizen science. Traditionally the main ethical challenges in citizen science have been identified as relating to data quality, data sharing and intellectual property, conflicts of interest, and the risk for exploitation of participants [10]. However, the distinguishing feature of personal science; that the person conducting the research is also the person being studied, sets it apart from most citizen science projects. Personal science projects are also in general of less of a collective nature than citizen science [11].

Examples of scientifically published personal science include Larry Smarr's self-diagnosis of inflammatory bowel disease from gut microbiome analyses [12] and Dana Lewis' work in type 1 diabetes, aiming to help both herself and the wider community by developing tools and methods to achieve improved blood glucose control [13]. It has been suggested that patients using personal science in collaboration with clinicians are in a better position to sustain a behavioural change [14].

In summary, the goal of personal science is not merely to collect data but rather to use self-collected data to achieve personally consequential insights that can be used for taking action in relation to a specific issue, often health related. Personal science is not intended to replace clinical research but rather to complement and enrich its practices and improve relevance to individual patients.

Personal science in PD

The practice of personal science has similarities to clinical N-of-1 studies, which have been used in PD by clinicians to study individuals with PD [15–17]. The key difference is that personal science is self-directed, meaning that the person conducting the study is also the person being studied. To the best of our knowledge, the only peer-reviewed academic work on personal science in PD has been conducted by the first author of this paper (SR); two single subject studies where SR used herself as the research participant [18,19]. The first study [18][6] was conducted with an observational design, exploring how the effects of SR's medication for PD, prescribed by her neurologist, varied across the day with time and with each medication intake. The medication effect was quantified by capturing finger tapping performance with a smartphone app. The second study [19][7] was conducted with a placebo-controlled interventional design, examining the effect of nicotine from an e-cigarette on levodopa-induced dyskinesias. In both studies, SR used the knowledge she gained to better understand her own personal condition and to improve treatment decisions, both with and without clinical support. In the following, the two personal science studies by SR will be used

6 Chapter 2 of this thesis
7 Chapter 3 of this thesis

to inform discussions around ethical aspect of personal science.

Ethical aspects of using self-tracking for scientific inquiry in PD

Group research is currently the cornerstone for implementing novel interventions into our healthcare systems and forms the basis for clinical guidelines and protocols and self-tracking as a method for data collection can be used also in that context. Wearable devices and other types of technology are proving to be useful tools for collecting data for research into PD at a group level, for example in studies using smartphone apps [20,21] or smartwatches [22], either alone or in combination with advanced clinical biomarkers, allowing for "deep phenotyping" [23].

However, the direct applicability of group research results to individual patients is limited and many of the personal questions that persons with PD have cannot be answered by group research. Examples of such unanswered questions that can be consequential on an individual level include: "How do I respond to this particular drug?"; "How can I time my medications to obtain the best possible effect?", "How can I find the best balance between functionality and medication side effects?", or "Do I sleep better when I exercise more?" This is where personal science can provide benefit.

In fact, the present discussion about personal science raises an almost philosophical issue about science in general, namely that the purpose of all research should ultimately be to benefit not the groups that were studied in a particular study, but rather individuals living with a chronic condition like PD. All too often, research findings are interpreted at the group level, without a sufficient understanding of the possible benefits (or harms) for the participating individuals. This issue is becoming all the more important as we are beginning to realise that PD is not a single condition with a single pathophysiology, but that it may be more appropriate to speak of 7 million different types of parkinsonism, namely as many as there are individuals living with this condition in the world [24]. And that we may ultimately need just as many personalised treatment approaches. The concept of personal science brings this approach a step closer to reality.

Of course, one could have a discussion about the adequacy of the term personal science, but that is beyond the scope of this paper. For our purposes it is enough to note that personal science can be seen as principally different from clinical research. We will reflect on these differences by elaborating on some important ethical considerations of using self-tracking for clinical and personal science respectively, focused on the topics: *self-tracking data*, *burden of tracking*, *relevance of research*, *independent review and dissemination,* and *protection and fair treatment of participants.*

Self-tracking data

When self-tracking is used for data collection in clinical research, the ethical responsibility lies with the clinician/researcher instructing the patient to collect data. When it comes to using data from digital tools in clinical research, clinical researchers are responsible for making sure that the privacy of the individuals generating the data is protected and potential risks mitigated [25]. As research into mHealth demonstrates, specific ethical issues can arise relating to patients' access to data, data ownership, privacy and security, and the potential exposure of bystanders [26].

Similarly, if digital tools are used to acquire data in personal science, privacy aspects can be an important issue. When individual patients use commercially available tools to track their own disease, there is an inherent risk that these health-related data may be exploited by private companies for their own purposes, such as targeted health advertisements. Of concern is also that such poorly protected health information finds its way to e.g. insurance companies, who may ultimately hold this against the participant by offering them a less attractive healthcare insurance policy. For individual patients, these long-term consequences are often not immediately apparent, and it may be more difficult for an individual to ascertain the privacy and security aspects when data are acquired with a particular commercially provided device. Such issues need to be addressed, including the question who can be held accountable for the potential risks of personal health data being handled by commercial tech companies.
In the two personal science studies by SR, data were collected using an app that saved data locally on the phone [18], as well as using pen and paper [18,19]. This demonstrates that even though ethics relating to self-tracking data can be an issue, personal science in PD can also be done without saving potentially sensitive data online.

7

Burden of tracking

Self-tracking can add a significant workload to the already demanding work of being a patient. In clinical group research, clinicians are obliged to minimise potential harm due to the intervention, and to make sure that any potential benefits outweigh the risk of harm to each individual participant [5]. Self-tracking may usefully alleviate the pressure on clinicians or researchers, but it certainly does not come "for free", as participants pay a price with their time investment, as demonstrated in previous research [27].
The aspect of added workload from self-tracking is especially important in PD, given the decreased energy levels and challenges with task management associated with PD. The two personal science studies in PD by SR specifically highlights this added burden of tracking [18,19].

Relevance of research

The questions explored in research have to be scientifically relevant while not exploiting

the participants [5]. This balance may be especially difficult to navigate in clinical group research for PD, since the time to potential benefit for both the individual person with PD participating in the clinical group research as well as to the wider community so far has largely failed to keep up with the speed of disease progression within an individual person with PD. Furthermore, the evidence is increasing that research priorities, as identified by persons with PD, often differ from priorities expressed by clinicians and researchers [28–30]. For example, clinicians tend to prioritise motor symptoms and other visible/quantifiable signs of PD, whereas many persons with PD lend greatest value to the less visible non-motor symptoms.

It is also worth noting that effects at the individual level can easily be lost at the group level perspective. For example, there are examples of drugs that have been approved based on research in groups that were dominated by men, even though in daily clinical practice, women prove to be much less responsive. Women may also experience significant side effects that were not seen in the study population dominated by men that participated in the original seeding trials. Such differences have also been observed in PD [31].

To address such issues, individualised research design, such as personal science, can provide benefit. From a relevance perspective, research into what persons with PD themselves consider important, using personal science, should be supported. Personal science has the potential to lead to insights that can inform further, more conventional systematic research and may thereby be able to contribute to improving the relevance of research. In personal science, the methodology (measurements, data collection, evaluation etc) is more likely to be generalisable, than the results. For example, in a highly variable and individualised condition like PD, an intervention that works for one person with PD might be unsuitable for another, for example because the efficacy or the risk of side effects can differ widely across different individuals. Being cautiously explicit about the limitations of the generalisability is a key element in sharing the results, which was done in the two articles by SR [18,19].

Independent review and dissemination

Independent review is important for ethical research to ensure that a researcher's conflicting interests do not cause problems, for example in the form of lower quality research [5]. In the US, independent review of clinical research is operationalised by for example granting agencies, local institutional review boards, and data and safety monitoring boards while other countries have other protective mechanisms. The structures for independent review can address different parts of the research process like study design, recruitment of participants etc. The peer reviewers and the journals' editors during the dissemination process can also be considered a form of independent

review. For clinical research, well-known procedures and safeguards are in place for all these phases.

For personal science the situation is currently unclear. Naturally, personal science practiced at the first level of impact, where the individual has the purpose to improve his/her own selfcare, is largely unproblematic. It is in particular when personal science is publicly disseminated that specific ethical challenges emerge. In situations like that, the transferability of the work conducted holds ethical implications since then the work can also have an effect beyond the individual performing the inquiry on themselves.
It has been argued that research led by patients requires adaptations of current ethical standards [32]. Should a person performing personal science with the intention to publish their findings somehow be protected from possibly harming themselves? This is an area where independent review could play an important role. A study of a group practicing personal science explored a process for joint ethical reflections and also present some suggested ethical principles, including transparency, participant control of data and ongoing risk-to-benefit evaluation [33]. We consider this among the most pressing issues regarding ethics of personal science; to explore appropriate mechanisms for independent review of personal science projects. Questions that need addressing include: How can independent review of personal science be implemented in a constructive manner so that new knowledge can be developed and disseminated without risk to personal scientists? At what stage is it reasonable to introduce mechanisms for protecting themselves from self-harm? How should the issue of informed consent be handled in personal science projects? Who should be responsible for deciding about the balance between safety and possible efficacy, as a regular ethics committee would normally do for group research? This area will require further work.

We have examined the two personal science studies in PD by SR with the issue of potential risk for self-harm in mind. For the observational study [18], the risk for self-harm can be considered low, since no other intervention than the medications prescribed by SR's neurologist was introduced. The interventional study [19] deserves more ethical attention, since it involves a self-chosen, non-medical intervention in the form of an e-cigarette. However, this specific intervention should be considered as being associated with a minimal additional risk, also in relation to the potential side effects that conventional medications for PD can entail.

When personal science projects are published in conventional scientific journals, established procedures apply, for example regarding ethical requirements. When SR's two studies [18,19] were published it was explicitly stated in the manuscripts that the studies had not been reviewed by an ethical review board and a description was given

as to how ethical aspects had been taken into account in conducting the study. Both studies were published after conventional review.

Protection and fair treatment of research participants
In clinical research studies selection of participants has to be done in a fair manner. This includes e.g. decisions on inclusion and exclusion criteria, recruitment strategies, study site selection, and populations to study [5]. The main difference between conventional clinical research and personal science is in the locus of control. Clinical research is conducted by clinicians on healthy participants and patients. Although conditions have improved as the terminology has evolved from *subjects* to *participants*, the fact remains that patients are typically not in control of the research process. Of course, a research participant has the right to discontinue their participation at any time, but then they will also miss out on any potential clinical benefits.

In personal science, the researcher and the participant are one and the same, and the primary goal for launching the personal science study is most often an explicit aim to gain a personal benefit, and this is a marked contrast to traditional group science. This means that in personal science, the participant/researcher is fully in control of the research process and can thereby decide in every stage of the project, if an invested effort is likely to yield sufficient benefits. These potential benefits can also lead to ethical challenges. For example, the desire to alleviate symptoms may motivate an individual to downplay the expected risks or effort associated with a certain intervention, which goes back to the discussion on protection against self-harm in the previous section.
It is also important from a value perspective that resources are used in a fair and just way. In general, patients doing personal science are individuals with high levels of autonomy [27]. They can pave the way for other patients but from an inclusivity perspective, it is important to realise that this route is not open for all. In further work on personal science, we must ensure that individuals and groups that are presently unable to engage in personal science for health, social, economic, or other reasons are not disadvantaged.

CONCLUSIONS AND FUTURE WORK

We conclude that current ethical requirements that are commonly applied to clinical group research, are not per se suitable for research conducted by persons with PD using personal science and that there is a need for development of adapted ethical procedures. To allow patient-led research in the form of personal science in PD to evolve further, specific ethical frameworks and regulations for self-tracking for personal science should be developed. The potential risk for self-inflicted harm should be

given specific attention. For a person wanting to engage in personal science projects, ethical aspects always have to be considered. In general, observational designs can be considered unproblematic. When personal science projects intended to be publicly disseminated use interventional design however, specific ethical challenges can arise, which may warrant independent ethical review. More work is needed in this field. A summary of key points and suggested future work is given in **Table 7.1**

TABLE 7.1: Summary of key points and future work.

Key points	
1.	The practice of personal science can evoke specific ethical challenges, in particular when personal science projects are publicly disseminated.
2.	Personal science projects using an observational design can generally be considered to raise fewer ethical challenges.
3.	For personal science projects using an interventional design that are performed with the intent to disseminate publicly, ethical challenges can arise relating to the risk for self-harm.
Future work	
1.	Specific ethical frameworks and regulations for personal science should be developed with a special focus on risks for self-harm, how to handle informed consent, and who should be responsible for decisions of the balance between safety and possible efficacy.
2.	Future work on personal science will need to include perspectives of diversity, inclusivity, and equitability.

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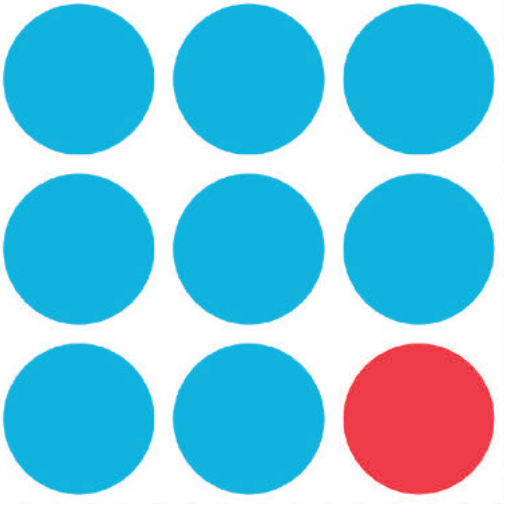
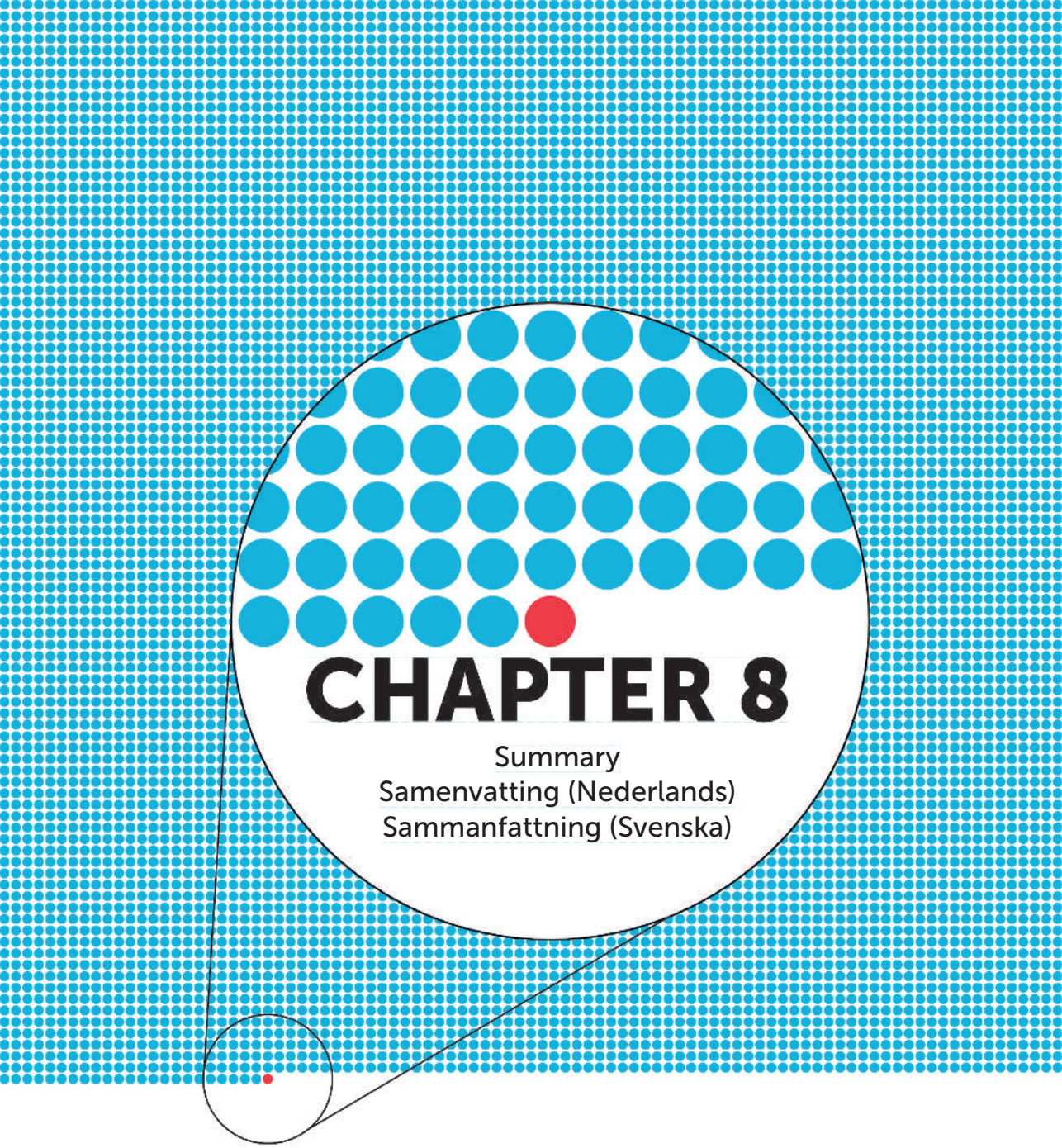
PART IV

SUMMARY AND GENERAL DISCUSSION



A while back, someone asked me: "What is the worst thing about having PD?". At first, I was surprised, as no one had ever asked me that before. When I had taken a few moments to go through the most frustrating aspects about this disease, I told the person: "The unpredictability of the disease, for example to wake up in the morning and not know what the day will be like, if my meds will kick in properly or if I will feel extra sluggish the whole day, if my walking today will be better or worse than usual. THAT is the worst thing about having PD!".



CHAPTER 8

Summary
Samenvatting (Nederlands)
Sammanfattning (Svenska)

SUMMARY

Parkinson's disease (PD) is a complex neurodegenerative condition with highly individualised manifestations and an extremely fluctuating nature. As the disease progresses, pharmaceutical treatments become increasingly complicated, and it becomes more and more challenging for persons with PD to manage their condition optimally. The burden falls increasingly on the persons living with this disease (and their family members) to get the combinations, doses, and timings of the multiple different medications right, and to find an optimal balance between the desired effects and the commonly occurring side effects. In addition, there is a wide range of nonpharmacological interventions – typically requiring involvement of a potentially very large set of healthcare professionals – that need to be integrated into the life of persons living with PD. And finally, lifestyle needs to be optimised, with a particular emphasis on regular exercise and a healthy diet. To date, persons with PD and their families are faced with this arduous task, largely without any tools for selfcare or with only relatively little support provided by the healthcare system. This process could be less complex if it were identical for each person living with PD, but the reality is that no two individuals are the same, so the one size fits all concepts do not apply to the management of PD. Consequently, effective selfcare is incredibly important for managing PD successfully. Adequate disease-specific knowledge is an important aspect of selfcare.

There is great potential for **patient-led research** to improve the relevance of selfcare research to persons with lived experience of diseases, including persons with PD. One type of patient-led research is **personal science**. As I outlined in **Chapter 1** and the **Glossary**, personal science is defined as *the practice of exploring personally consequential questions by conducting self-directed N-of-1 studies using a structured empirical approach*. Personal science is a framework of study and includes **self-tracking** as a main method for data collection. Self-tracking is defined as *collecting and structuring observations about one's own life* and can also be used for collecting data in conventional clinical research. Also mentioned in **Chapter 1** is that self-tracking (both technology based, and non-technology based) has the potential to improve selfcare. Although the complexity of PD makes self-tracking particularly challenging, it is plausible to assume that many persons with PD would benefit from a better understanding of the possibilities of self-tracking in the context of personal science. Therefore, this thesis has examined how patient-led research in the form of personal science might contribute to selfcare in PD.

Part I: Personal science in PD – exploring feasibility

In **Part I** of this thesis, I explored how personal science can be used to further facilitate selfcare in PD. This was done by using a single subject research design with myself

as both the researcher as well as the research subject. This thesis contains two personal science studies, one with an observational design (**Chapter 2**) and one with an interventional design (**Chapter 3**).

In this thesis, I use the term personal science to refer to the overarching framework of self-directed N-of-1 studies. The two studies/chapters use slightly different terminology. Here, for the sake of clarity, the term *personal science* will be used to mean both *patient-initiated self-tracking* (term used in **Chapter 2**) and *patient-driven N-of-1 studies* (term used in **Chapter 3**).

Using an observational design (**Chapter 2**), my main objective was to explore personal science as a means to assess the effects of my medication and how they vary on a daily basis. The observed phenomenon that functioned as a primary outcome measure of the medication effect was my finger tapping ability. Data were collected over four subsequent days using technology (an app on my smartphone) as well as with pen and paper. Only data from the first two days were complete enough to analyse, leading to the realisation that the collection of data over a period of time can pose a significant burden to persons engaging in personal science. I also realised that my motor function was not the same throughout the day. Specifically, I observed a dip in finger function around the time for the second medication dose of each day.

Personal science can, furthermore, be used to evaluate the individual effects of an intervention, for example evaluating how to best manage the side effects of medication. For the interventional design in this thesis (**Chapter 3**), the main objective was to explore the potential role for personal science as a method for improving selfcare in PD using the example of managing a troublesome side effect of PD medication, namely levodopa-induced dyskinesia (LID), using nicotine as a therapeutic intervention. The observations in **Chapter 3** were recorded using pen and paper and the studied phenomenon was LID. The results indicate that nicotine administered via an e-cigarette may possibly have an effect on LID in an individual person with PD, although a placebo effect could also have explained the observed improvement that I noted in my own symptoms. The main contribution of this study is, however, highlighting the work done by patients on a daily basis for understanding their conditions and conducting personal science.

I found that interacting with the collected data and their visualisation during the analysis and created feedback loops contributing to my learning. It led to me learning about both the specifics of my different medications as well as what the combination of them means for my motor function over the course of the day. When data from personal science is shared during a clinical encounter, it potentially has the double benefit of contributing to the learning of both patient and clinician.

The practice of personal science for PD is facilitated by having a better knowledge of PD (including relevant symptoms, available treatments, and potential side effects), by gaining some experience in data collection and visualisation, by learning skills in problem solving, and by having a positive attitude. This kind of knowledge and skill can be difficult to achieve and may take time to acquire, yet is essential if personal science is to produce meaningful insights. Explicit support from healthcare professionals, family members or friends, and peer groups or individuals is likely to be beneficial in this regard.

The single subject studies in this thesis (**Chapter 2 & 3**) demonstrate the potential value of personal science. The main proposition from these two studies is that using personal science as a framework for self-observation and symptom tracking has the potential to enable persons with PD to better understand their own personal condition and to improve their treatment, both with and without support from clinicians. Importantly, I found that the *burden of tracking*, meaning the burden added by the process of self-tracking, was more demanding than expected. Even when the study is designed and conducted by the person being studied, there are limitations to what a participant in a single case research study wants to do.

Part II: Personal science in PD – transferability of the methods of personal science
In **Part II**, I investigated the needs, abilities, and attitudes of persons with PD other than myself, in relation to the methods tested in **Part I.** Specific questions included: are the needs of other persons with PD in Sweden relating to their selfcare similar to mine (as I found in **Part I**)? Can they be expected to have, or be able to acquire, the knowledge and abilities necessary to engage in personal science? Do they also have the interest in, and attitudes supportive of, making use of discoveries acquired through personal science?

The current state of selfcare for PD in Sweden was explored in **Chapter 4**. The aims of this study were twofold. First, to investigate how persons with PD acquire disease-specific knowledge, meaning all knowledge relating to PD, including but not limited to knowledge about symptoms, medication and other treatments, side effects, disease management and healthcare provision. Second, to assess to what extent persons with PD use PD-related healthcare services.

Data were collected during four weeks in March-April 2015 through an online survey which had 346 respondents. Of these, 51% were men, the age range was 16-87 years (median age 68 years), and the time since diagnosis varied from recently diagnosed to 31 years (median 7 years). The chosen online format meant that the response percentage could not be calculated, and that any response bias could not be formally

ascertained. The age and gender distributions were, however, consistent with what would be expected for the overall population of Swedish persons with PD. A large majority of the respondents (91%) considered knowledge about PD to be important and about half of the respondents (55%) reported to have been able to acquire the knowledge they needed. When asked about their main source of disease-specific knowledge, 36% responded that they had found the information themselves online, 29% from patient organisations, and 25% from their healthcare providers.

When it comes to the time spent on healthcare-related issues, 8% (n=29) of our respondents did not see a neurologist at all during the year studied, and 60% (n=206) saw their neurologist for up to an hour. Only 47% had regular contact with other healthcare professionals for health issues related to their PD. Among our respondents, 43% considered the time they saw their neurologist to be sufficient, while 35% thought it was insufficient, and 22% were neutral.

The self-reported level of disease-specific knowledge was not significantly correlated with gender, age (under/over 65), education level, time since diagnosis (shorter/longer than 7 years), or time spent with the neurologist (estimated from the self-reported numbers of visits and self-reported average length of time per visit). The effect of the presence of a Parkinson nurse was not investigated.

Main sources of disease-specific knowledge were not significantly correlated with education level, time since diagnosis, or time spent with the neurologist. However, significantly different sources were reported depending on age and gender. Online information was the main source of disease-specific knowledge for persons with PD under 65 years of age (online: 45%) whereas the group of persons of 65 years and older more often reported patient organisations as their main source (online: 31%). Women reported finding their information more often online (online: 40%) compared to men (online: 31%).

As expected, there was a significant association between spending more time per year with their treating neurologist and being satisfied with the amount of time spent. However, the satisfaction of persons with PD with the time they receive with their neurologist was not significantly associated with age, gender, time since diagnosis or education level.

A significant association was found between self-reported level of knowledge and satisfaction with the time with the neurologist every year. This could imply that persons with PD reporting a higher level of knowledge were more likely to be satisfied with the time they had with their neurologist, regardless of how short or long that time

was. It might also indicate that patients get more out of their consultation with the neurologist if they arrive at the scene better prepared, i.e. with more disease-specific knowledge, so that the depth and quality of the discussion is improved.

Experiences and opinions of persons with PD in Sweden on personal science were investigated in **Chapter 5**. In the study in **Chapter 5**, the term self-tracking should be read to mean personal science. A mixed methods' approach was used to generate a model for personal science in PD. Qualitative data from seven interviews were combined with quantitative data from a survey. As for the survey in Chapter 4, an online format was used, which meant that response percentages could not be calculated. The survey was online between December 7, 2017, and January 7, 2018. In total, 280 persons with PD responded to the survey, 64% (n = 180) of whom had experience with personal science. Of these, 51% were men, average age was 64.4 years, and average time since diagnosis was 7.7 years.

The results indicate that it can be both rewarding and challenging to make discoveries through personal science. Different persons with PD track different aspects of his or her disease and treatment, for example timings and/or types of medication, and exercise or physical activity. Different individuals may also use different methodologies to track their disease. Mostly they track only in their head, but sometimes they also make notes on paper or use technology. The findings in **Chapter 5** confirm the results from **Part I** concerning the burden of tracking. Persons with PD consider tracking to require a lot of work and the right individual balance between burdens and benefits needs to be found. The main identified benefits are that personal science gives persons with PD a deeper understanding of their own specific manifestations of PD and contributes to a more effective decision making regarding their own selfcare. They see personal science as a method they can use to better understand and actively manage their PD. Furthermore, personal science is considered to be able to contribute to personalised healthcare.

8

The main identified burdens of tracking include emphasising the patient role and that it can be difficult to know what and how to track. Also, the respondents strongly believe that their healthcare providers need better ways to assess PD. Furthermore, they do see a risk of becoming obsessed with tracking and think it is important that self-tracking does not begin to dominate their lives.

Some strategies that people use in order to achieve this optimal balance have been identified. A first possible solution was to focus on positive aspects rather than negative ones. One of the interviewees emphasised the importance of finding the right phenomenon to observe in relation to the question explored. The person had started

out registering pain and stiffness but soon realised that to focus on such negative aspects did not encourage continued tracking. The person chose to turn it around and started tracking "feel well time", i.e. when the PD symptoms were reasonably well under control, which improved motivation to continue tracking.

A second possible solution was to track specific aspects only occasionally, for example to help understand how medications relate to symptom relief and side effects and to remind themselves of how their disease varies over time.

The third strategy was to increase the benefits by interacting with the data collected, thereby creating feedback loops to enable persons with PD to learn from their own data.

Part III: Patient-led research in PD

In **Part III**, I examined some aspects of patient-led research. In **Chapter 6,** some opportunities and challenges of patient-led research and patient researchers were described. In **Chapter 7,** I compared ethical aspects of using self-tracking for personal science to using self-tracking in the context of conducting clinical research on groups of study participants.

In **Chapter 6**, I briefly described the different roles that patients can take in research, the passive research participant, the partly active role of research partners, and the active and often largely autonomous role of patient researchers. There is great potential for patient researchers in contributing to improving biomedical research since they can bridge the lived and the learned experiences of diseases. I conclude that the contributions of patient researchers should be formally recognised, that patients interested in becoming patient researchers should be encouraged and supported, and that healthcare professionals and scientists without lived experience should be educated to appreciate patient researchers' contributions.

In **Chapter 7**, ethical aspects of different roles for patients in research are discussed using the example of self-tracking. Self-tracking is a method for data collection that can be used in conventional clinical group research (research participants) and in personal science (patient researchers). Conventional group-based clinical research aims to find generalisable answers to clinical or public health questions. The aim of personal science is different: to find meaningful answers that matter first and foremost to an individual with a particular health challenge. When persons with chronic diseases use empirical methods to improve their own selfcare and start to deploy the possibilities of the Internet and other technological developments to conduct their own research, specific ethical challenges can emerge. One such issue can arise when individuals also

intend to disseminate their findings. Sharing can occur for example by publishing in a scientific journal, or via posts on social media. As was discussed in **Chapter 7**, the researcher and the participant are one and the same in the framework of personal science, and there is a need for development of adapted ethical procedures. To allow patient-led research in the form of personal science in PD to evolve further, specific ethical frameworks and regulations for self-tracking for personal science should be developed. The potential risk for self-inflicted harm should be given specific attention.

SAMENVATTING (NEDERLANDS)

De Ziekte van Parkinson (Parkinson's Disease, PD) is een complexe neurodegeneratieve aandoening met sterk geïndividualiseerde verschijnselen en een uiterst fluctuerend karakter. Naarmate de ziekte voortschrijdt, worden de farmaceutische behandelingen steeds ingewikkelder en wordt het voor personen met PD steeds moeilijker om hun aandoening optimaal te managen. Het wordt steeds moeilijker voor mensen met de ziekte (en hun familieleden) om de juiste combinaties, doseringen en tijdstippen van de vele verschillende geneesmiddelen te vinden en een optimaal evenwicht te vinden tussen de gewenste effecten en de vaak optredende bijwerkingen. Daarnaast is er een breed scala aan niet-farmacologische interventies - die meestal de betrokkenheid van een potentieel zeer groot aantal zorgverleners vereisen - die moeten worden geïntegreerd in het leven van mensen die leven met de PD. Ten slotte moet de levensstijl worden geoptimaliseerd, met bijzondere nadruk op regelmatige lichaamsbeweging en gezonde voeding. Tot op heden staan personen met PD en hun families alleen voor deze zware taak, grotendeels zonder hulpmiddelen voor zelfzorg of met slechts relatief weinig steun van het zorgstelsel. Dit proces zou minder complex kunnen zijn als het identiek zou zijn voor elke persoon met PD, maar de realiteit is dat geen twee individuen hetzelfde zijn, dus de "one size fits all" concepten zijn niet van toepassing op het management van PD. Daarom is effectieve zelfzorg ongelooflijk belangrijk voor het succesvol omgaan met PD. Adequate ziektespecifieke kennis is een belangrijk aspect van zelfzorg.

Er is een groot potentieel voor patiënt-gestuurd onderzoek om de relevantie van zelfzorg onderzoek te verbeteren voor mensen die een ziekte aan den lijve ondervinden, waaronder mensen met PD. Eén type van patiënt-gestuurd onderzoek is persoonlijke wetenschap. Zoals ik in Hoofdstuk 1 en de Verklarende Woordenlijst heb uiteengezet, wordt persoonlijke wetenschap gedefinieerd als de praktijk van het onderzoeken van persoonlijk consequentiële vragen door het uitvoeren van zelf-gerichte N-of-1 studies met behulp van een gestructureerde empirische benadering. Personal science/ persoonlijke wetenschap is een studiekader en omvat self-tracking als belangrijkste methode voor dataverzameling. Self-tracking wordt gedefinieerd als het verzamelen en structureren van observaties over het eigen leven en kan ook worden gebruikt voor het verzamelen van gegevens in conventioneel klinisch onderzoek. Ook wordt in hoofdstuk 1 vermeld dat self-tracking (zowel gebaseerd op technologie, als op niet-technologie) de potentie heeft om zelfzorg te verbeteren. Hoewel de complexiteit van PD self-tracking bijzonder uitdagend maakt, is het aannemelijk te veronderstellen dat veel personen met PD baat zouden hebben bij een beter begrip van de mogelijkheden van self-tracking in de context van persoonlijke wetenschap.

Daarom is in dit proefschrift onderzocht hoe patiënt-gestuurd onderzoek in de vorm

van personal science/persoonlijke wetenschap zou kunnen bijdragen aan zelfzorg bij PD.

Deel I: Persoonlijke wetenschap bij PD - verkenning van de haalbaarheid
In deel I van dit proefschrift heb ik onderzocht hoe personal science gebruikt kan worden om zelfzorg bij PD verder te faciliteren. Dit heb ik gedaan door gebruik te maken van een single subject onderzoeksopzet met mijzelf als onderzoeker, zowel als onderzoekssubject. Dit proefschrift bevat twee onderzoeken naar persoonlijke wetenschap, één met een observationeel ontwerp (Hoofdstuk 2) en één met een interventioneel ontwerp (Hoofdstuk 3).

In dit proefschrift gebruik ik de term/het begrip persoonlijke wetenschap om te verwijzen naar het overkoepelende raamwerk van zelfgestuurde N-van-1/N=1 studies. De twee studies/hoofdstukken gebruiken enigszins verschillende terminologie. Hier, voor de duidelijkheid, zal de term/het begrip personal science gebruikt worden om zowel patiënt-geïnitieerde self-tracking (term gebruikt in Hoofdstuk 2) als patiënt-gestuurde N-of-1 studies (term gebruikt in Hoofdstuk 3) aan te duiden.

Gebruikmakend van een observationeel design (Hoofdstuk 2), was het mijn mijn hoofddoel om personal science te onderzoeken als een middel om de effecten van mijn medicatie te beoordelen en hoe ik deze zou kunnen variëren op een dagelijkse basis. Het geobserveerde fenomeen dat fungeerde als een primaire maat voor de uitkomst van het medicatie-effect was mijn vingertikvaardigheid . De gegevens werden gedurende vier opeenvolgende dagen verzameld met behulp van technologie (een app op mijn smartphone) en met pen en papier. Alleen de gegevens van de eerste twee dagen waren compleet genoeg om te analyseren, wat leidde tot het besef dat het verzamelen van gegevens over een bepaalde periode een aanzienlijke belasting kan vormen voor wie zich bezighoudt met persoonlijke wetenschap. Ik realiseerde me ook dat mijn motoriek niet de hele dag hetzelfde was. Ik zag met name een dip in de vingerfunctie rond het tijdstip van de tweede medicatiedosis van elke dag.

Persoonlijke wetenschap kan bovendien gebruikt worden om de individuele effecten van een interventie te evalueren, bijvoorbeeld om te evalueren hoe het beste omgegaan kan worden met de bijwerkingen van medicatie. Voor het interventionele ontwerp in dit proefschrift (Hoofdstuk 3) was het hoofddoel om de potentiële rol van personal science als methode voor het verbeteren van zelfzorg bij PD te onderzoeken aan de hand van het voorbeeld van het beheersen van een lastige bijwerking van PD medicatie, namelijk levodopa-geïnduceerde dyskinesie (LID), met behulp van nicotine als een therapeutische interventie. De observaties in hoofdstuk 3 zijn genoteerd met pen en papier en het bestudeerde fenomeen was LID. De resultaten geven aan dat

nicotine toegediend via een e-sigaret mogelijk een effect heeft op LID in een individueel persoon met PD, hoewel een placebo-effect ook de waargenomen verbetering, die ik in mijn eigen symptomen heb waargenomen, zou kunnen hebben verklaard. De belangrijkste bijdrage van dit onderzoek is echter dat het de aandacht vestigt op het werk dat patiënten dagelijks doen om hun aandoeningen te begrijpen en persoonlijke wetenschap te bedrijven.

Ik constateerde dat de interactie met de verzamelde gegevens en de visualisatie ervan tijdens de analyse en feedback loops creëerden die bijdroegen aan mijn leerproces. Het leidde ertoe dat ik zowel de specifieke kenmerken van mijn verschillende medicaties leerde kennen als de betekenis van de combinatie ervan voor mijn motorische functie in de loop van de dag. Wanneer gegevens van persoonlijke wetenschap worden gedeeld tijdens een klinische ontmoeting, heeft dit het dubbele voordeel dat het bijdraagt aan het leren door zowel de patiënt als de clinicus/arts.

Het beoefenen van persoonlijke wetenschap voor PD wordt vergemakkelijkt door een betere kennis van PD (inclusief relevante symptomen, beschikbare behandelingen, en mogelijke bijwerkingen), door enige ervaring op te doen met het verzamelen en visualiseren van gegevens, door vaardigheden te leren in het oplossen van problemen en door een positieve houding te hebben. Het kan tijd en moeite kosten om dit soort kennis en vaardigheden te verkrijgen, maar de kennis en vaardigheden zijn essentieel als persoonlijke wetenschap tot zinvolle inzichten moet leiden. Expliciete steun van professionals in de gezondheidszorg, familieleden of vrienden en peer groups of individuen is waarschijnlijk gunstig in dit opzicht.

De single subject studies in dit proefschrift (Hoofdstuk 2 & 3) tonen de potentiële waarde van personal science aan. De belangrijkste stelling uit deze twee studies is dat het gebruik van persoonlijke wetenschap als een kader voor zelfobservatie en het bijhouden van symptomen het potentieel heeft om mensen met Parkinson in staat te stellen hun eigen persoonlijke conditie beter te begrijpen en hun behandeling te verbeteren, zowel met als zonder ondersteuning van specialisten. Belangrijk is dat ik ontdekte dat de last van het volgen, d.w.z. de last die wordt toegevoegd door het proces van self-tracking, zwaarder was dan verwacht. Zelfs wanneer de studie ontworpen en uitgevoerd wordt door de persoon die bestudeerd wordt, zijn er beperkingen aan de inspanning die een deelnemer aa een single case research studie wil leveren . Natuurlijk is het ook moeilijk om vooraf te begrijpen hoeveel belasting men kan verwachten.

Deel II: Persoonlijke wetenschap bij Parkinson - overdraagbaarheid van de methoden van persoonlijke wetenschap

In deel II onderzocht ik de behoeften, bekwaamheden en attitudes van andere personen

met PD dan ikzelf, in relatie tot de in deel I geteste methoden. Specifieke vragen waren onder meer: zijn de behoeften van andere personen met PD in Zweden met betrekking tot hun zelfzorg vergelijkbaar met de mijne (zoals ik in deel I constateerde)? Kan van hen verwacht worden dat ze de kennis en vaardigheden hebben, of kunnen verwerven, die nodig is om aan persoonlijke wetenschap te doen? Hebben zij ook belangstelling voor het benutten van ontdekkingen die zij via persoonlijke wetenschap hebben opgedaan? Hebben ze de attitude die daarvoor bevordelijk is?

De huidige stand van zaken van zelfzorg voor PD in Zweden werd onderzocht in Hoofdstuk 4. De doelstelling van deze studie was tweeledig. Ten eerste, te onderzoeken hoe personen met PD ziektespecifieke kennis verwerven, d.w.z. alle kennis met betrekking tot PD, inclusief, maar niet beperkt tot kennis van symptomen, medicatie en andere behandelingen, bijwerkingen, ziektemanagement en zorgverlening. Ten tweede, na te gaan in welke mate personen met PD gebruik maken van PD-gerelateerde gezondheidszorgdiensten/zorgaanbod.

De gegevens werden verzameld over een periode van van 4 weken in maart en april 2015 via een online enquête die 346 respondenten telde. Hiervan was 51% man, de leeftijd varieerde van 16-87 jaar (mediaan 68 jaar) en de tijd sinds de diagnose varieerde van recent gediagnosticeerd tot 31 jaar geleden gediagnosticeerd (mediaan 7 jaar). Door de gekozen online opzet kon het responspercentage niet worden berekend en kon een eventuele vertekening van de respons niet formeel worden vastgesteld. De verdeling naar leeftijd en geslacht kwam echter overeen met wat kan worden verwacht voor de totale populatie van Zweedse PD-patiënten. Een grote meerderheid van de respondenten (91%) vond kennis over PD belangrijk en ongeveer de helft van de respondenten (55%) gaf aan dat zij de nodige kennis hadden kunnen verwerven. Op de vraag naar hun belangrijkste bron van ziektespecifieke kennis antwoordde 36% dat zij de informatie zelf online hadden gevonden, 29% bij patiëntenorganisaties, en 25% van hun zorgverleners.

Wat de tijd betreft die aan zorggerelateerde zaken werd besteed, zag 8% (n=29) van onze respondenten helemaal geen neuroloog tijdens het bestudeerde jaar en 60% (n=206) zag hun neuroloog gedurende maximaal een uur. Slechts 47% had regelmatig contact met andere zorgverleners voor gezondheidszaken die verband hadden met hun Parkinson.. Van onze respondenten vond 43% de hoeveelheid tijd die zij hun neuroloog zagen voldoende, terwijl 35% het onvoldoende vond en 22% neutraal was. Het zelfgerapporteerde niveau van ziektespecifieke kennis was niet significant gecorreleerd met geslacht, leeftijd (jonger/ ouder dan 65), opleidingsniveau, tijd sinds diagnose (korter/langer dan 7 jaar), of tijd doorgebracht bij de neuroloog (geschat op basis van het zelfgerapporteerde aantal bezoeken en de zelfgerapporteerde

gemiddelde tijdsduur per bezoek). Het effect van de aanwezigheid van een Parkinson verpleegkundige werd niet onderzocht.

De belangrijkste bronnen van ziektespecifieke kennis waren niet significant gecorreleerd met opleidingsniveau, tijd sinds de diagnose, of tijd doorgebracht bij de neuroloog. Er werden echter significant verschillende bronnen gerapporteerd afhankelijk van leeftijd en geslacht. Online informatie was de belangrijkste bron van ziektespecifieke kennis voor personen met PD jonger dan 65 jaar (online: 45%), terwijl de groep van personen van 65 jaar en ouder vaker patiëntenorganisaties als hun belangrijkste bron opgaven (online: 31%). Vrouwen gaven vaker aan hun informatie online te vinden (online: 40%) in vergelijking tot mannen (online: 31%).

Zoals verwacht was er een significant verband tussen het aantal contacturen of minuten met de behandelend neuroloog en de mate van tevredenheid met het aantal contactminuten/contacturen. Echter, de tevredenheid van personen met PD met de tijd die zij krijgen van hun neuroloog was niet significant geassocieerd met leeftijd, geslacht, tijd sinds diagnose of opleidingsniveau.

Er werd een significant verband gevonden tussen het zelfgerapporteerde kennisniveau en de tevredenheid met de tijd/het aantal contacturen of minuten die ze jaarlijks bij de neuroloog doorbrengen. Dit zou kunnen impliceren dat personen met PD die een hoger kennisniveau rapporteerden meer kans hadden om tevreden te zijn met de tijd die ze met hun neuroloog hadden, ongeacht hoe kort of lang die tijd was. Het zou er ook op kunnen wijzen dat patiënten meer uit hun consult met de neuroloog halen als zij beter voorbereid, dat wil zeggen met meer ziektespecifieke kennis, ter plaatse komen, zodat de diepgang en kwaliteit van het gesprek wordt verbeterd.

Ervaringen en meningen van personen met PD in Zweden over persoonlijke wetenschap werden onderzocht in Hoofdstuk 5. In het onderzoek in Hoofdstuk 5 moet de term self-tracking gelezen worden als persoonlijke wetenschap. Een 'mixed methods' benadering werd gebruikt om een model voor persoonlijke wetenschap bij PD te genereren. Kwalitatieve gegevens uit zeven interviews werden gecombineerd met kwantitatieve gegevens uit een enquête. Net als voor de enquête in hoofdstuk 4 werd een online format gebruikt, waardoor responspercentages niet konden worden berekend. De enquête stond online van 7 december 2017 tot 7 januari 2018. In totaal reageerden 280 personen met PD op de enquête, van wie 64% (n = 180) ervaring had met persoonlijke wetenschap. Van hen was 51% man, de gemiddelde leeftijd was 64,4 jaar en de gemiddelde tijd sinds de diagnose was 7,7 jaar.

De resultaten geven aan dat het zowel belonend als uitdagend kan zijn om

ontdekkingen te doen via persoonlijke wetenschap. Verschillende personen met PD volgen verschillende aspecten van zijn of haar ziekte en behandeling, bijvoorbeeld tijdstippen en/of soorten medicatie en lichaamsbeweging of fysieke activiteit. Verschillende personen kunnen ook verschillende methodologieën gebruiken om hun ziekte te volgen. Meestal houden ze het alleen in hun hoofd bij, maar soms maken ze ook aantekeningen op papier of gebruiken ze technologie. De bevindingen in hoofdstuk 5 bevestigen de resultaten uit deel I met betrekking tot de belasting van het bijhouden. Personen met PD vinden dat het bijhouden van gegevens veel werk met zich meebrengt en ze geven aan dat er een goed individueel evenwicht moet worden gevonden tussen de lasten en de voordelen. De belangrijkste geconstateerde voordelen zijn dat persoonlijke wetenschap personen met PD een dieper inzicht geeft in hun eigen specifieke manifestaties van PD en dat het bijdraagt aan een effectievere besluitvorming met betrekking tot zelfzorg. Zij zien persoonlijke wetenschap als een methode om hun PD beter te begrijpen en er actief mee om te gaan. Bovendien verwachten de respondenten dat persoonlijke wetenschap kan bedragen aan gepersonaliseerde gezondheidszorg.

De belangrijkste geïdentificeerde lasten van tracking zijn onder andere het benadrukken van de rol van de patiënt en dat het moeilijk kan zijn om te weten wat en hoe te tracken. Ook zijn de respondenten sterk van mening dat hun zorgverleners betere methodes moeten hebben hebben om het verloop van hun Parkinson te beoordelen. Verder zien ze een risico van geobsedeerd raken door self-tracking en vinden ze het belangrijk dat self-tracking hun leven niet gaat domineren.

Er zijn enkele strategieën geïdentificeerd die mensen gebruiken om het optimale evenwicht tussen de lasten en voordelen van self-tracking te proberen te bereiken. Een eerste mogelijke oplossing was om de nadruk te leggen op positieve aspecten in plaats van negatieve. Eén van de geïnterviewden benadrukte het belang van het vinden van het juiste Parkinson verschijnsel/Parkinson fenomeen om te observeren in relatie tot de onderzochte vraag. De persoon was begonnen met het registreren van pijn en stijfheid, maar realiseerde zich al snel dat het focussen op zulke negatieve aspecten niet bevorderlijk was voor het verder volgen. De persoon koos ervoor om het om te draaien en begon met het bijhouden van “feel well time”, d.w.z. wanneer de PD-symptomen redelijk goed onder controle waren, wat de motivatie om door te gaan met het bijhouden verbeterde.

Een tweede mogelijke oplossing was het slechts af en toe volgen van specifieke aspecten, bijvoorbeeld om te helpen begrijpen hoe medicijnen verband houden met verlichting van symptomen en bijwerkingen en om zichzelf eraan te herinneren hoe hun de verschijnselen van hun ziekte in de loop van de tijd varieëren.

De derde strategie bestond uit het vergroten van de voordelen door interactie met de verzamelde gegevens, waardoor feedback loops ontstonden, waarmee mensen met Parkinson van hun eigen gegevens kunnen leren.

Deel III: Door de patiënt geleid onderzoek bij PD
In deel III heb ik enkele aspecten van patiënt-gestuurd onderzoek onderzocht. In hoofdstuk 6 werden enkele kansen en uitdagingen van patiëntgestuurd onderzoek en patiëntonderzoekers beschreven. In Hoofdstuk 7 vergeleek ik ethische aspecten van het gebruik van self-tracking voor persoonlijke wetenschap met het gebruik van self-tracking in de context van het uitvoeren van klinisch onderzoek bij groepen studiedeelnemers.

In hoofdstuk 6 beschreef ik kort de verschillende rollen die patiënten in onderzoek kunnen innemen, de passieve onderzoeksdeelnemer, de deels actieve rol van onderzoekspartners, en de actieve, vaak grotendeels autonome rol van patiëntonderzoekers. Er ligt een groot potentieel voor patiëntonderzoekers om bij te dragen aan de verbetering van biomedisch onderzoek, aangezien zij een brug kunnen slaan tussen de doorleefde en de geleerde ervaringen met ziekten. Ik concludeer dat de bijdragen van patiëntonderzoekers formeel moeten worden erkend, dat patiënten die geïnteresseerd zijn in de rol van patiëntonderzoeker moeten worden aangemoedigd en ondersteund en dat zorgverleners en wetenschappers zonder doorleefde ervaring moeten worden opgeleid om de bijdragen van patiëntonderzoekers te waarderen.

I conclude that the contributions of patient researchers should be formally recognised, that patients interested in becoming patient researchers should be encouraged and supported, and that healthcare professionals and scientists without lived experience should be educated to appreciate patient researchers' contributions.

In **Hoofdstuk 7** worden de ethische aspecten van verschillende rollen voor patiënten in onderzoek besproken aan de hand van het voorbeeld van self-tracking. Self-tracking is een methode voor dataverzameling die kan worden gebruikt in conventioneel klinisch groepsonderzoek (onderzoeksdeelnemers) en in persoonlijke wetenschap (patiëntonderzoekers). Conventioneel klinisch onderzoek in groepsverband heeft tot doel generaliseerbare antwoorden te vinden op klinische of volksgezondheidsvragen. Het doel van persoonlijke wetenschap is anders: het vinden van zinvolle antwoorden die in de eerste plaats van belang zijn voor een individu met een bepaalde gezondheidsuitdaging. Wanneer personen met chronische ziekten empirische methoden gebruiken om hun eigen zelfzorg te verbeteren en de mogelijkheden van het internet en andere technologische ontwikkelingen beginnen te benutten om hun eigen onderzoek te verrichten, kunnen specifieke ethische uitdagingen ontstaan. Een van die

problemen kan zich voordoen wanneer personen ook van plan zijn hun bevindingen te verspreiden. Het delen kan bijvoorbeeld gebeuren door hun bevindingen te publiceren in een wetenschappelijk tijdschrift, of via posts op sociale media. Zoals in **Hoofdstuk 7** is besproken, zijn de onderzoeker en de deelnemer een en dezelfde in het kader van persoonsgebonden wetenschap, en is er behoefte aan de ontwikkeling van aangepaste ethische procedures. Om patiëntgestuurd onderzoek in de vorm van personal science bij PD verder te laten evolueren, moeten specifieke ethische kaders en regelgeving voor self-tracking voor personal science worden ontwikkeld. Het potentiële risico op zelf toegebrachte schade moet specifieke aandacht krijgen.

SAMMANFATTNING (SVENSKA)

Parkinsons sjukdom är en komplex neurodegenerativ sjukdom med mycket individuella manifestationer och en extremt fluktuerande karaktär. I takt med att sjukdomen fortskrider blir läkemedelsbehandlingen alltmer komplicerad, och det blir allt svårare att hantera sjukdomen optimalt. Det faller alltmer på de personer som lever med denna sjukdom (och deras familjemedlemmar) att hitta rätt kombinationer, doser och tidpunkter för de många olika läkemedlen och att hitta en optimal balans mellan de önskade effekterna och biverkningarna. Dessutom finns det ett stort antal icke-farmakologiska åtgärder - som vanligtvis kräver medverkan av flera olika vårdprofessioner - som måste integreras i det dagliga livet för personer som lever med Parkinson. Slutligen måste livsstilen optimeras, med särskild tonvikt på regelbunden motion och hälsosam kost. Hittills har personer med Parkinson och deras familjer fått hantera denna svåra uppgift i stort sett helt utan verktyg för egenvård eller med relativt begränsat stöd från hälso- och sjukvårdssystemet. Denna process skulle kunna vara mindre komplicerad om den var identisk för varje person som lever med Parkinson, men i realiteten är ingen individ den andra lik. Det finns därför ingen standard som passar alla när det gäller behandling av Parkinson. En effektiv egenvård är därmed otroligt viktigt för att hantera Parkinson på ett framgångsrikt sätt. Sjukdomsspecifik kunskap är en viktig aspekt av egenvården.

Det finns en stor potential för **patientledd forskning** att förbättra relevansen av forskning om egenvård för personer med levd erfarenhet av sjukdomar, inklusive personer med Parkinson. En typ av patientledd forskning är **"personal science"**. Som jag beskrev i **kapitel 1** och i **ordlistan** så definieras personal science som *en metod för att utforska frågor av stor personlig vikt genom att genomföra självstyrda N-of-1-studier med hjälp av ett strukturerat empiriskt tillvägagångssätt*. Personal science är ett ramverk och den viktigaste metoden för datainsamling är egenmonitorering ("self-tracking" på engelska). Egenmonitorering innebär insamling och strukturering av observationer om det egna livet och kan även användas för insamling av data inom konventionell klinisk forskning. I **kapitel 1** nämns också att egenmonitorering (både teknikbaserade och icke teknikbaserade) har potential att förbättra egenvården. Även om egenmonitoreringar i Parkinson försvåras av sjukdomens komplexitet, är det rimligt att anta att många personer med Parkinson skulle gynnas av en bättre förståelse av möjligheterna med egenmonitorering och personal science.

I denna avhandling har jag därför undersökt hur patientledd forskning i form av personal science kan bidra till egenvård för Parkinson.

Del I: Personal science för Parkinson - undersökning av genomförbarheten

I **del I** av denna avhandling har jag undersökt hur personal science kan användas för att förbättra egenvården för Parkinsons sjukdom. Detta gjorde jag genom att använda mig själv som forskningsperson. Denna avhandling innehåller två personal science-studier, en designad som en observationsstudie (**kapitel 2**) och den andra som en interventionsstudie (**kapitel 3**).

I den här avhandlingen använder jag termen personal science som beteckning för självstyrda N-of-1-studier. I de två studierna/kapitlen i denna del används något olika terminologi. För tydlighetens skull kommer termen personal science här att användas för att beteckna både patientinitierad egenmonitorering (term som används i **kapitel 2**) och patientdrivna N-of-1-studier (term som används i **kapitel 3**).

I observationsstudien (**kapitel 2**) var syftet att använda personal science för att utforska effekterna av min medicinering och variationer över dagen. Jag använde min snabbhet i att knacka med fingrarna på en smartphone som mått på medicineringseffekten. Data samlades in under fyra på varandra följande dagar med hjälp av teknik (en app på min smartphone) samt med penna och papper. Endast data från de två första dagarna var tillräckligt fullständiga för att kunna analyseras, vilket ledde till insikten att insamling av data under en längre tid kan utgöra en betydande börda för personer som ägnar sig åt personal science. Jag insåg också att min motoriska funktion inte var densamma under hela dagen. Specifikt observerade jag en sämre fingerfunktion runt tiden för den andra läkemedelsdosen varje dag.

Personal science kan även användas för att utvärdera de individuella effekterna av en åtgärd, t.ex. för att bättre förstå läkemedelsbiverkningar. För interventionsstudien i denna avhandling (**kapitel 3**) var huvudsyftet att utforska personal science som en metod för att förbättra egenvård vid Parkinson. Detta gjordes genom att använda personal science för att hantera en besvärlig biverkning av parkinsonmedicinering, nämligen levodopa-inducerad dyskinesi (LID), med hjälp av nikotin som en terapeutisk intervention. Observationerna i **kapitel 3** registrerades med penna och papper och det studerade fenomenet var LID. Resultaten tyder på att nikotin som administreras via en e-cigarett möjligen kan ha en effekt på LID hos en enskild person med Parkinson, även om en placeboeffekt också skulle kunna ha förklarat den observerade förbättringen som jag noterade i mina egna symtom. Det viktigaste bidraget från denna studie är dock att belysa det arbete som patienter gör varje dag för att förstå sina sjukdomar och bedriva personal science.

Jag fann att när jag interagerade med mina insamlade observationer och visualiserade dem under analysen så skapades återkoppling som bidrog till mitt eget lärande. Det

ledde till att jag lärde mig både om specifika detaljer i mina olika mediciner och hur kombinationen av dem påverkar min motoriska funktion under dagens lopp. Om resultat från personal science också diskuteras under läkarbesök så kan det ha den dubbla fördelen att bidra till lärandet för både patient och läkare.

Möjligheterna att tillämpa personal science för Parkinson förbättras om man har mer kunskaper om Parkinson (inklusive relevanta symtom, tillgängliga behandlingar och potentiella biverkningar), om man har viss erfarenhet av datainsamling och visualisering, om man lär sig strukturerad problemlösning och om man har en positiv attityd. Denna typ av kunskaper och färdigheter kan vara svåra att uppnå och kan ta tid att förvärva, men är viktiga om personal science ska ge meningsfulla insikter. Stöd från vårdpersonal, familjemedlemmar eller vänner samt andra patienter kan vara till nytta i detta avseende.

De första två studierna i denna avhandling (**kapitel 2 och 3**) visar på det potentiella värdet av personal science. Den viktigaste insikten från dessa två studier är att användningen av personal science kan användas för egna observationer och symtomuppföljning och kan därmed göra det möjligt för personer med PD att bättre förstå sin egen situation och att förbättra sin behandling, både med och utan stöd från vårdpersonal. Viktigt är även att jag fann att ”mätningsbördan”, det vill säga den börda som tillkommer av arbetet med att göra egenmonitorering, var mer krävande än väntat. Detta innebär att även när studien utformas och genomförs av den person som studerar sig själv, finns det begränsningar för hur mycket arbete en deltagare i en forskningsstudie är beredd att ta på sig.

Del II: Personal science för Parkinson – metodernas överförbarhet

I **del II** undersökte jag behoven, förmågorna och attityderna relaterat till de metoder som testades i **del I**, hos andra personer med Parkinson än jag själv. Specifika frågor var bland annat: Är behoven hos andra personer med Parkinson i Sverige liknande mina (som jag fann i **del I**) när det gäller deras egenvård? Kan man förvänta sig att de har, eller kan förvärva, de kunskaper och förmågor som krävs för att ägna sig åt personal science? Har de också det intresse och de attityder som stödjer upptäckter genom personal science?

I **kapitel 4** undersöktes läget när det gäller egenvård för Parkinson i Sverige. Studien hade två syften. För det första att undersöka hur personer med Parkinson förvärvar sjukdomsspecifik kunskap, det vill säga all kunskap som rör Parkinson, inklusive, men inte begränsat till, kunskap om symtom, medicinering och andra behandlingar, biverkningar, sjukdomshantering och sjukvård. För det andra att bedöma i vilken utsträckning personer med Parkinson använder parkinsonrelaterade hälso- och

sjukvårdstjänster.

Data samlades in under fyra veckor i mars-april 2015 med hjälp av en onlineenkät med 346 respondenter. Av dessa var 51 % män, åldersintervallet var 16-87 år (medianålder 68 år) och tiden sedan diagnosen varierade från nyligen diagnostiserad till 31 år (median 7 år). Det valda onlineformatet innebar att svarsprocenten inte kunde beräknas. Ålders- och könsfördelningen överensstämde dock med vad som kan förväntas för den svenska populationen av personer med Parkinson. En stor majoritet av de svarande (91 %) ansåg att kunskap om Parkinson är viktig och ungefär hälften av de svarande (55 %) rapporterade att de hade kunnat skaffa sig den kunskap de behövde. På frågan om deras främsta källa till sjukdomsspecifik kunskap svarade 36 % att de själva hade hittat informationen på nätet, 29 % från patientorganisationer och 25 % från sin vårdgivare.

När det gäller tid i vården så träffade 8 % (n=29) av våra respondenter inte en neurolog alls under det aktuella året, och 60 % (n=206) träffade sin neurolog i upp till en timme. Endast 47 % hade regelbunden kontakt med annan sjukvårdspersonal för hälsofrågor som rörde deras Parkinson. Bland våra respondenter ansåg 43 % att den tid de träffade sin neurolog var tillräcklig, medan 35 % ansåg att den var otillräcklig och 22 % var neutrala.

Den självrapporterade nivån av sjukdomsspecifik kunskap var inte signifikant korrelerad med kön, ålder (under/över 65 år), utbildningsnivå, tid sedan diagnosen (kortare/längre än 7 år) eller tid som tillbringats hos neurologen (uppskattad från det självrapporteringen av antalet besök och den genomsnittliga tiden per besök). Effekten av tillgång till en Parkinsonsjuksköterska undersöktes inte.

De viktigaste källorna till sjukdomsspecifik kunskap var inte signifikant korrelerade med utbildningsnivå, tid sedan diagnosen eller tid hos neurologen. Olika källor rapporterades dock beroende på ålder och kön. Information på nätet var den viktigaste källan till sjukdomsspecifik kunskap för personer med Parkinson under 65 år (på nätet: 45 %, från vården: 21 %, från patientorganisationer: 19 %, övriga källor: 15 %), medan gruppen av personer som var 65 år och äldre oftare rapporterade patientorganisationer som sin viktigaste källa (på nätet: 31 %, från vården: 27 %, från patientorganisationer: 33 %, övriga källor: 8 %). Kvinnor rapporterade att de oftare hittade sin information online (på nätet: 40 %, från vården: 18 %, från patientorganisationer: 32 %, övriga källor: 10 %) jämfört med män (på nätet: 31 %, från vården: 32 %, från patientorganisationer: 26 %, övriga källor: 11 %).

Som väntat fanns det ett signifikant samband mellan mer tid per år hos sin behandlande neurolog och att vara nöjd med den tiden. Hur nöjda personer med Parkinson var med

den tid de får med sin neurolog hade dock inget signifikant samband med ålder, kön, tid sedan diagnosen eller utbildningsnivå.

Ett signifikant samband hittades mellan självrapporterad kunskapsnivå och hur nöjd man var med tiden hos neurologen varje år. Detta skulle kunna innebära att personer med Parkinson som rapporterade en högre kunskapsnivå hade större sannolikhet att vara nöjda med den tid de fick med sin neurolog, oavsett hur kort eller lång den tiden var. Det skulle också kunna tyda på att patienterna får ut mer av sitt möte med neurologen om de kommer dit bättre förberedda, dvs. med mer sjukdomsspecifik kunskap, så att diskussionens djup och kvalitet förbättras.

Erfarenheter och åsikter hos personer med Parkinson i Sverige om personal science undersöktes i **kapitel 5**. I studien i **kapitel 5** ska begreppet self-tracking läsas som personal science. En ansats med blandade metoder användes för att ta fram en modell för personal science för Parkinson. Kvalitativa data från sju intervjuer kombinerades med kvantitativa data från en enkät. Liksom för enkäten i **kapitel 4** användes ett onlineformat, vilket innebar att svarsprocenten inte kunde beräknas. Enkäten var öppen mellan 7 december 2017 och 7 januari 2018. Totalt svarade 280 personer med Parkinson på enkäten, varav 64 % (n = 180) hade erfarenhet av personal science. Av dessa var 51 % män, medelåldern var 64,4 år och den genomsnittliga tiden sedan diagnosen var 7,7 år.

Resultaten visar att det kan vara både givande och utmanande att göra upptäckter genom personal science. Olika personer med Parkinson följer olika aspekter av sin sjukdom och behandling, t.ex. tidpunkter och/eller typer av medicinering samt motion eller fysisk aktivitet. Olika personer kan också använda olika metoder för att följa sin sjukdom. Oftast håller de koll bara i huvudet, men ibland gör de också anteckningar på papper eller använder teknik. Resultaten i **kapitel 5** bekräftar resultaten från **del I** när det gäller mätningsbördan. Personer med Parkinson anser att egenmonitorering kräver mycket arbete och att man behöver hitta den rätta individuella balansen mellan bördor och fördelar. De viktigaste identifierade fördelarna är att personal science kan ge personer med Parkinson en djupare förståelse för deras egna specifika yttringar av Parkinson och kan bidra till ett effektivare beslutsfattande om egenvården. De ser personal science som en metod som de kan använda för att bättre förstå och aktivt hantera sin sjukdom. Personal science anses även kunna bidra till en mer personcentrerad hälso- och sjukvård.

De viktigaste identifierade bördorna är att personal science kan betona patientrollen och att det kan vara svårt att veta vad och hur man ska spåra. Respondenterna anser också att deras vårdgivare behöver bättre sätt att bedöma Parkinson. De anser att det

finns en risk för att bli fixerad av egenmonitoreringen och att det är viktigt att de inte börjar dominera deras liv.

Några strategier som människor använder för att uppnå denna optimala balans har identifierats. En första möjlig lösning var att fokusera på positiva aspekter snarare än negativa. En av de intervjuade betonade vikten av att hitta rätt fenomen att observera i förhållande till den undersökta frågan. Personen hade börjat med att registrera smärta och stelhet men insåg snart att fokusering på sådana negativa aspekter inte uppmuntrade till fortsatta mätningar. Personen valde därför att vända på saken och började registrera "må bra-tiden", dvs. när parkinsonsymtomen var någorlunda väl under kontroll, vilket ökade motivationen för att fortsätta registreringen.

En annan möjlig lösning var att endast mäta specifika aspekter under en begränsad tid, t.ex. för att hjälpa till att förstå hur mediciner relaterar till symtomlindring och biverkningar och för att påminna sig själv om hur sjukdomen varierar med tiden.

Den tredje strategin var att öka nyttan genom att interagera med insamlad data och på så sätt skapa återkoppling som gör det möjligt för personer med Parkinson att lära sig av sina egna observationer.

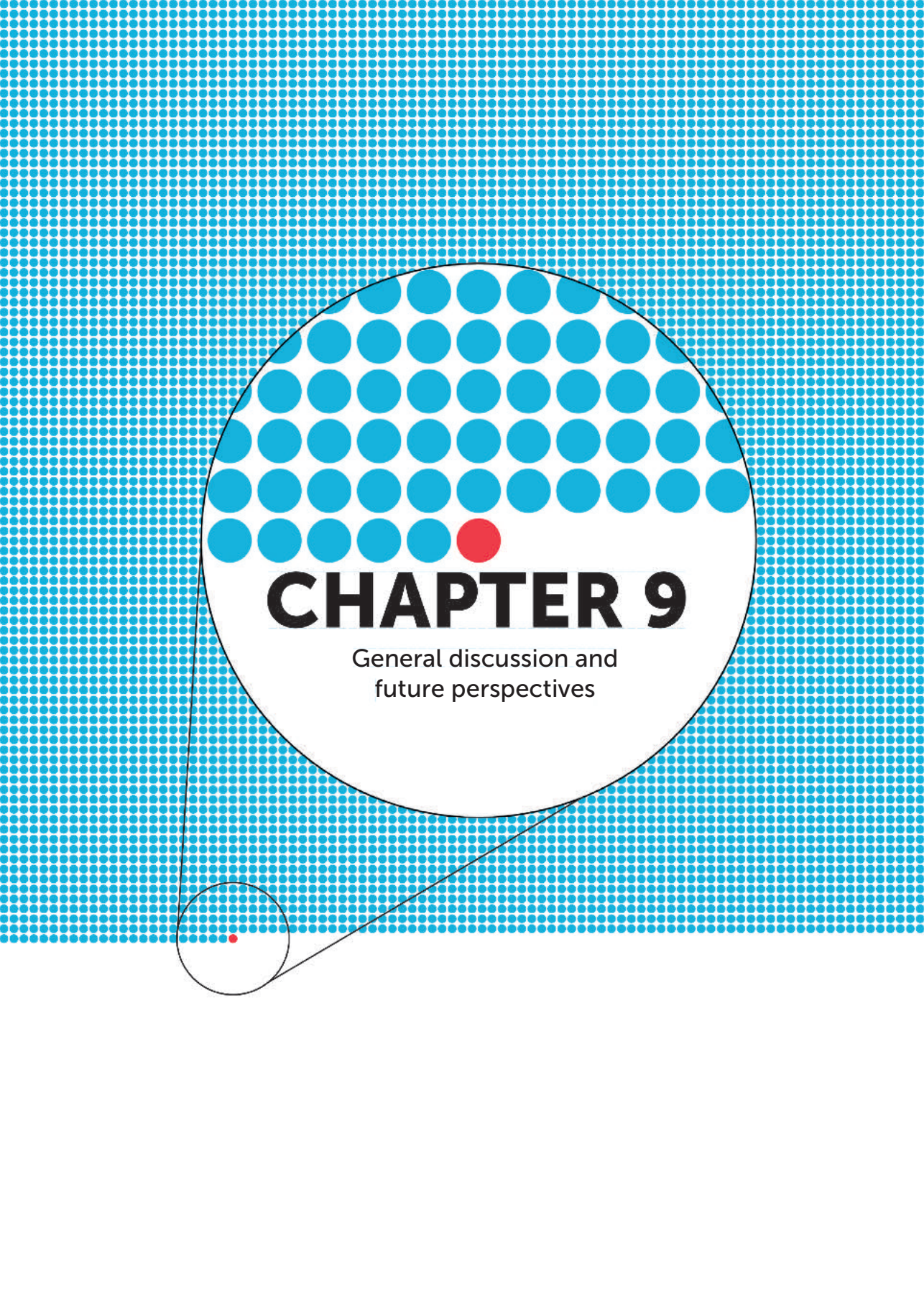
Del III: Patientledd forskning i Parkinson

I **del III** undersökte jag aspekter av patientledd forskning. I **kapitel 6** beskrevs vissa möjligheter och utmaningar med patientledd forskning och patientforskare. I **kapitel 7** jämförde jag etiska aspekter av att använda egenmonitorering för personal science med att använda egenmonitorering i samband med klinisk forskning på grupper av studiedeltagare.

I **kapitel 6** beskrev jag kortfattat de olika roller som patienter kan ta i forskning; den passiva forskningsdeltagaren, den delvis aktiva rollen som forskningspartner och den aktiva och ofta till stor del självständiga rollen som patientforskare. Det finns en stor potential för patientforskare när det gäller att bidra till att förbättra den biomedicinska forskningen, eftersom de kan överbrygga de levda och lärda erfarenheterna av sjukdomar. Jag drar slutsatsen att patientforskarnas bidrag bör erkännas formellt, att patienter som är intresserade av att bli patientforskare bör uppmuntras och stödjas, och att vårdpersonal och forskare som inte har erfarenhet av patientforskning bör utbildas för att kunna uppskatta patientforskarnas bidrag.

I **kapitel 7** diskuteras etiska aspekter av olika roller för patienter i forskning med hjälp av exemplet egenmonitorering. Egenmonitorering är en metod för datainsamling som kan användas inom konventionell klinisk gruppforskning (forskningsdeltagare) och

inom personal science (patientforskare). Konventionell gruppbaserad klinisk forskning syftar till att finna generaliserbara svar på kliniska frågor eller frågor om folkhälsa. Syftet med personlig forskning är annorlunda: att hitta meningsfulla svar som först och främst är viktiga för en individ med en särskild hälsoutmaning. När personer med kroniska sjukdomar använder empiriska metoder för att förbättra sin egenvård och börjar utnyttja de möjligheter som Internet och annan teknisk utveckling erbjuder för att bedriva sin egen forskning, kan särskilda etiska utmaningar uppstå. En sådan fråga kan uppstå när enskilda personer också har för avsikt att sprida sina upptäckter. Spridning kan till exempel ske genom att publicera i en vetenskaplig tidskrift eller via inlägg på sociala medier. Som diskuterades i **kapitel 7** är forskaren och deltagaren en och samma person inom ramen för personal science, och det finns ett behov av att utveckla anpassade etiska regelverk. För att patientledd forskning i form av personal science för Parkinson ska kunna utvecklas vidare bör särskilda etiska ramar och regler för egenmonitorering inom personal science utvecklas. Den potentiella risken för självskadebeteende bör ägnas särskild uppmärksamhet.



CHAPTER 9

General discussion and
future perspectives

GENERAL DISCUSSION

The overall aim of this thesis was to examine how **patient-led research** in the form of **personal science** can contribute to selfcare for persons living with Parkinson's disease. In this thesis, **patient-led research** is defined as research led by **patient researchers**; persons with lived experience of a disease, disability, or other health challenge who are openly using those experiences in doing research, within academia or in other contexts. **Personal science** is a framework of study defined as: *"the practice of exploring personally consequential questions by conducting self-directed N-of-1 studies using a structured empirical approach"* and it is known to be used regarding health and well-being.

Parkinson's disease (PD) has been known to medical science for over two centuries. During that time, therapeutic advances have transformed it from a fatal condition to a disease that can be managed with variable success in the long term [1]. For example, there now exist a wide array of pharmacological as well as nonpharmacological interventions, including surgical treatments and multidisciplinary care. However, currently available treatments are not able to halt this very complex and individually varying disease from progressing, resulting, over time, in a substantial reduction of quality of life.

I would argue that medical science to date has failed to provide persons with PD with useful means for managing the full complexity of PD on a daily basis and tailored to each individual's unique personal needs, *i.e.,* to equip persons with PD with the means to engage in effective selfcare. In this thesis, I have demonstrated that patient-led research in the form of personal science can contribute to improving selfcare in PD. These elements will be further elaborated on in this chapter, where I will also provide suggestions for future directions. I have chosen to present the discussion in three main sections: i) Practical issues are covered in the section *How can personal science be used to improve selfcare in PD?*, ii) Philosophical reasoning around barriers and how these can be overcome are covered in the section *How can patient-led research and personal science reach their full potential?*, and iii) Future directions and recommendations are given, for academics, clinicians, and persons with PD.

How can personal science be used to improve selfcare in PD?

"Know thyself"
- Temple of Apollo at Delphi

Living with PD is challenging, and it can be very difficult to balance effects and side effects of the many different available therapeutic options to find the optimal regimen.

Symptoms and the response to treatment can vary greatly across different times of the day for the same individual, and may also differ widely between different days. Even the seasons can be a source of further fluctuations [2]. Since PD is a progressive disorder (which means that existing symptoms worsen over time, and that new symptoms may arise), in time it is associated with increasingly complex selfcare. The absence of objective biomarkers makes selfcare even more challenging. Persons with PD have to manage their condition every day of the year, which includes regulating a complex regime of medication intakes including different combinations of different drugs and finding the right timings. As an illustration of the treatment complexity persons with PD can face, an example of my daily intake of PD medications can be seen in **Figure 9.1**. And the complexity of the daily management process is further compounded by the presence of a wide variety of nonpharmacological interventions and lifestyle advice.
From the citations referred to in **Chapter 1**, it is clear that there is extensive research demonstrating the importance of persons with PD being engaged and knowledgeable. Nevertheless, selfcare is an under-researched field, not only in PD but more in general as well. This goes for both theoretical knowledge, e.g., what the different elements of a successful selfcare regime for PD would be, as well as practical knowledge, e.g., what kind of selfcare support different persons with PD would need and favour.

FIGURE 9.1: Example of my daily intake of PD medications with planned timings. My regimen consists of five different prescription drugs taken at seven times each day in different combinations.

Using personal science to improve selfcare

In this thesis, my focus is on how personal science can be used to improve selfcare in PD. One common misunderstanding is to think that personal science is the same as self-tracking. As it is used in this thesis, self-tracking is a method for collecting data that can be used in personal science as well as in conventional clinical research. In both cases, data are collected by an individual about different aspects of that person's life and health. The main difference is that in conventional research, data are collected to respond to questions posed by a clinical researcher, most often relating to group level concerns. In personal science, both the questions asked, and the data collected are in the control of the individual.

In this thesis, I have demonstrated that personal science has the potential to improve selfcare in PD for a single subject (**Chapters 2 & 3**). Furthermore, I have provided some first evidence that the methods of personal science may be transferable to a wider population of persons with PD in Sweden (**Chapter 5**). In this latter chapter, I cannot exclude the possibility of a skewed selection. This means that it is possible that the respondents to my survey represented a subgroup of more highly educated, cognitively intact and non-depressed persons with PD. It is well known that certain subpopulations, for example persons with lower education levels or those from non-Western origin, are much more difficult to reach with online surveys. Efforts to reach out to such groups should continue to be high on the research agenda. Nevertheless, I feel that is reasonable to conclude that a sizeable proportion of persons with PD in Sweden have the autonomy and abilities needed for personal science as well as the attitudes and interests necessary to benefit. It is, however, also reasonable to assume that not all persons with PD will be able to benefit from personal science practices. One direction for future research is to explore how one can predict which individuals are likely to benefit and which are not. As I emphasised before, such an approach cannot be successful if issues around diversity are not addressed adequately. This will involve a proactive outreach to traditionally underserved populations, so that they can begin to improve from selfcare and personal science as well.

The studies in this thesis do give some insights into the question of which persons with PD could be more interested in personal science. Women and men seem to have slightly different approaches: compared to men, women are more likely to find their knowledge about PD online (**Chapter 4**) and are also more likely to track specific aspects (**Chapter 5**) and they track to prepare for the upcoming doctor's appointments (**Chapter 5**). A person's age also seems to affect their behaviour in this field: persons with PD younger than 65 are more likely to find their PD specific knowledge online, whereas persons with PD older than 65 are more likely to report patient organisations as their main source of knowledge (**Chapter 4**). Furthermore, persons with PD younger

than 65 are more likely to have a positive attitude towards and use tracking than older persons with PD (**Chapter 5**). Interestingly, the source of knowledge about PD did not appear to be affected by the time passed since the diagnosis was established (**Chapter 4**). In the study on attitudes and experiences of personal science, however, I found that persons with PD diagnosed more than 5 years ago were more likely to use tracking to understand their PD as well as more likely to use their tracking results together with their physician than persons with PD diagnosed less than 5 years ago (**Chapter 5**). Taken together, this could potentially be interpreted to mean that the ones most likely to be open towards and interested in personal science among persons with PD could be women younger than 65 who have been diagnosed for more than 5 years. This obviously needs to be confirmed in further studies. Having this type of knowledge is important, as it could help to define a proactive outreach to subgroups of patients who would be less inclined to resort to personal science, aiming to improve their ability to participate in selfcare as well.

Engaging in personal science requires a broader skillset than usual for persons with PD. It requires knowledge about available treatments and their possible (side)effects, as well as skills to use assessment methods, innovative technologies, and data analyses. These conclusions are confirmed in research from other areas [3–6]. Even though it is likely that not all persons with PD will have the necessary skills, knowledge, and attitudes, **Chapters 4** and **5** suggest that many persons with PD have the ability to potentially benefit from personal science. I therefore conclude that the merits of personal science likely extend beyond just me as an individual. A broader adoption of the practices of personal science will however most likely require dedicated training and education of both patients and healthcare professionals, since many persons with PD are likely to need more support to optimally implement selfcare. Worth noting is that engaging in personal science is always voluntary, so an individual always has the right not to engage. One argument to discard self-tracking could be a perceived burden, as I will discuss in more detail below. A further discussion on the future perspectives can also be found below.

Another common misunderstanding is to think that personal science is mainly about collecting and sharing personal data. In fact, as research within the Quantified Self community has demonstrated, sharing methods can be more important than sharing data [5,7,8]. Furthermore, as I show in **Chapter 2, 3 & 5**, the insights do not primarily come from the collection of data itself, but rather from the interaction with your data and the opportunity to learn from them.

Perhaps unsurprisingly, this thesis suggests that persons with PD with more knowledge about PD are more autonomous. Similar to my own experience (see cover image),

many persons with PD see their neurologist for only a rather short time each year. The results of **Chapter 4** show that 68% of respondents saw their neurologist for one hour per year or less. While no significant association was found between the amount of time with the neurologist and the level or source of knowledge, persons with PD reporting a higher level of knowledge were more likely to be more satisfied with the amount of time they had with their neurologist, regardless of how short or long that time was. There is also the possibility that patients are more satisfied with the quality of their interaction with the neurologist if they arrive at the consultation better prepared, i.e. with more disease-specific knowledge, so that the depth and quality of the discussion is improved.

As described in **Chapter 7**, personal science can be practiced at different levels of impact: at the first level, I used personal science to improve my own selfcare and at level 2, I also communicated the methods used as well as my findings publicly (**Chapters 2 & 3**). I chose to do so by publishing my studies in scientific journals but as mentioned in **Chapter 1**, personal science projects can also be made public through blogs, presentations, videos or similar. Whether or not personal scientists have a moral obligation to publish their findings, just like traditional scientists do [9], is an interesting matter of debate, that I address in further detail in **Chapter 7**.

Benefits and burdens

The benefits of using personal science include enabling persons with PD to gain a deeper understanding of, and actively managing, their PD, improving selfcare, and contributing to personalised medicine (**Chapters 2, 3 and 5**). This is also graphically illustrated in **Figure 9.2**.

It is important to note that self-tracking does not come for free. The findings in **Chapters 2, 3, and 5** suggest that the process of tracking is regulated by a balance between benefits and burdens. This means that the benefits, as perceived by the person who is tracking, have to be larger than the expected burdens. These findings are consistent with research in other research areas, such as type 1 diabetes [10,11] and mixed/multiple chronic conditions [12,13]. Similar conclusions are also known from the field of PD [14,15]. The burden of tracking has several dimensions, see **Figure 9.2**. First, self-tracking can be perceived as "illness work" [12,14], adding tasks to a person's everyday life (**Chapters 2, 3 & 5**), thereby emphasising the patient role. These tasks take time and energy, that has to be taken away from other activities, which might be activities that the person actually prefers. This includes specific challenges related to PD, which is a condition that is known to affect both the physical and mental abilities of affected individuals. One well-known consequence is the common difficulty of persons with PD to effectively engage in multitasking, or to oversee complex daily

schedules [16]. These disease features can make it overwhelming for persons with PD to also engage in frequent self-tracking. This burden also includes that it can be demoralising to track a degenerative condition, knowing that you are tracking an inevitable decline. It can be frustrating for patients to benchmark themselves against other fellow patients, because one might for example notice that your own disease progression is more rapid than that of others, and this could cause understandable anxiety and stress.

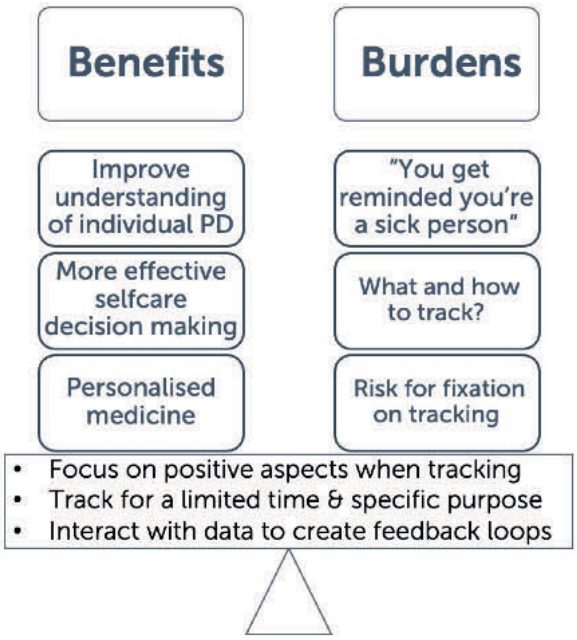


FIGURE 9.2: Balancing benefits and burdens of personal science through the use of balancing strategies. Benefits in the left stack, burdens in the right stack, and balancing strategies under the stacks.

Second, it can be difficult to know how and what to track (**Chapter 5**). Which data to collect and for whose sake. One reason for self-tracking can be that a healthcare provider has asked a person to track specific parameters. This can lead to problems, since healthcare providers often do not understand the individually lived experience of chronic disease, as suggested by examples from type 1 diabetes [10,11] and PD [15]. The issue of how to track can also lead to frustration. Should one use simple and straightforward pen and paper, or perhaps more advanced technology? The latter approach may provide a more detailed insight, but can also be much more challenging

to manage. Costs may also play a role here, because many individuals who engage in personal science use the smartphone and/or smartwatch which they had purchased themselves. But not everybody is able to possess such technologies.

The third identified burden was that persons with PD in the study in **Chapter 5** saw a risk for fixating on tracking. This particular burden can be specific to PD, since exaggerated internet use can be sign of impulse control disorders in PD [17]. Excessive use of tracking devices could be seen as "digital punding" (unnecessary stereotypical behaviours).

A few potential ways to reduce the burden of tracking (**Figure 9.2**) are to: focus on positive aspects ("feel-well-time", see **Chapter 5**), track for a specific purpose and for a limited time (see **Chapters 2, 3, and 5**), and interact with the collected data to create feedback loops that can contribute to learning (see **Chapters 2, 3, and 5**). These findings are supported by research conducted in other fields [12,18,19].

The role of technology in personal science for PD

> *"Not everything that can be counted counts,*
> *and not everything that counts can be counted."*
> *- William Bruce Cameron, sociologist (1963)*

Many initiatives and projects in recent years have used technology to track different aspects of PD [20–22]. To date, only a few are generally available, and then often only to clinicians, which suggests that they have been developed primarily to meet the needs of healthcare providers rather than to support persons with PD in their selfcare. Below, I will present a few examples of technologies that have been developed for use in PD and discuss them in relation to personal science. The examples are chosen as illustrations and are not intended to represent a comprehensive review of the use of technology in PD.

The Personal Kinetigraph (PKG) [23] is a smartwatch, i.e. worn on the wrist, collecting data on movements (tremor, bradykinesia, and dyskinesia) and providing reminders for medication intakes. The REMPARK/STAT-ON system [24,25] consists of a sensor worn in a belt which records the movements of persons with PD. For both of these devices, the data collected by persons with PD are not available to the individual generating the data. This implies that they are designed to support the needs of healthcare professionals rather than to support selfcare of persons with PD.

A project taking a more patient-centred approach is the smartphone app mPower. It was launched as part of the Apple Research Kit in March 2015 with the aim of developing

a better understanding of the variations of PD and their potential modulators, as well as for providing real-time feedback to the participants [26]. Although the mPower app was initially downloaded 48,104 times, only 0.3% (n=150) of users were persons with PD who contributed data for five separate days or more [27]. Data were collected for a number of activities and tasks, and persons with PD were given feedback in the form of their voice score and number of finger taps completed [28]. As discussed in **Chapter 1**, it has been demonstrated that providing personalised feedback to persons with PD based on individually collected data can improve health and selfcare [29–31]. Unfortunately, it is not possible to assess what the effects on persons with PD were when they received this feedback based on their mPower data because no evaluation of that kind has been done [28]. One potential explanation for the high attrition rates of mPower users could be the lack of support provided for selfcare for persons with PD in the versions of the app released to date. Another reason might be that the episodic completion of the tasks at home was not engaging enough, or perhaps it had to be done too frequently. Research from various areas has shown that the use of gamification elements helps to improve engagement with remote interventions and assessments [12,32,33], so perhaps this is something that could be considered for future versions. Finally, it might be that the data were not automatically coupled to the primary healthcare process, so that it could become part of the discussion between patient and healthcare provider in the consultation room. So far, mPower has been used for research purposes only and it is unclear if as part of further developments, it is intended to primarily address the needs of healthcare professionals or if it will also be used to support persons with PD in their selfcare.

There are some important differences between how I work to improve my selfcare in PD and the examples of technologies mentioned above. First, I chose myself what phenomenon of PD I wanted to study and how. This means that I could focus on the aspects of my PD that were of specific concern and interest to me, like variations in the medication effect throughout the day (**Chapter 2**) or the management of troublesome side effects (**Chapter 3**). It also means that I could use technology where it added the greatest value, and not use technology simply for the sake of using technology. The study in **Chapter 2** benefited from using technology (a smartphone app) for collecting data on finger tapping performance, whereas the data collection in **Chapter 3** was done using pen and paper. This gives me the freedom to work adaptively and balance the burdens and benefits of the self-tracking work.

Second, the use of data is very different. In the three examples above (PKG, REMPARK/STAT-ON, and mPower), data collected by an individual are interpreted using algorithms developed for a group level interpretation of how PD manifests. In my studies (**Chapters 2 & 3**), the data that I generated were very personal and full of

meaning to me, and I use them to compare the results only with those of myself at different times. This means that the data are probably not of interest to anyone else. It also means that if algorithms developed for a group level had been used to interpret my data, the individual meaning to me would likely have been lost.

Third, from managing my PD for over 35 years, I have learned the importance of details. I know that on some days, for no apparent reason, my medications just will not give the effect that I expect and that I am used to. I also know that there are a lot of factors influencing my PD and also the effect of my medications; a few examples are sleep patterns, exercise, stress, food, and emotional state. This is also a well-known phenomenon for other persons with PD, as the study in **Chapter 5** demonstrates; many persons with PD know that their response to their PD medications, as well as their physical and cognitive functionality are strongly influenced by factors such as a bad night's sleep, stress levels, a concurrent infection etc. In the studies that I have conducted on myself, I can choose to record and consider context of this kind, whereas in conventional group studies, details like these are rarely captured. The main reason for that is probably because it would mean a completely unreasonable workload for persons living with PD. But for me, making notes also of the context of my data enables me to both learn and adapt better than I otherwise could.

As for the experiences of other persons with PD, **Chapter 5** shows that the most commonly tracked elements were related to medication intake times, medication types, and physical activity and exercise. Most respondents used pen and paper solutions (56%) and one in two persons with PD (49%) had at some time used technology for their self-tracking.

In conclusion, technology does have potential for improving the lives of persons with PD, but the way new solutions are developed needs to change [34]. As I have demonstrated in **Chapters 2, 3 & 5**, the most important feature of personal science does not lie in the tools or technology used but in the fact that the individuals who are exploring their own questions are in control of the self-tracking process. For technology to be of best use for selfcare for persons with PD, the development process has to start with the needs of persons with PD rather than the needs of healthcare professionals or researchers.

It is important to note that the role of technology in personal science for other conditions/diseases can be very different. For conditions with clear objective biomarkers, technology can potentially be a game changer. It is, however, worth emphasising that pen and paper can also be very powerful tracking tools.

Especially when personal science is done with technical devices such as smartphones or wearables like smartwatches or other activity trackers, issues relating to data integrity, data quality as well as privacy and security issues come into play. Different brands of devices use different algorithms for interpreting the data collected. This means that for example the resulting number of steps in a day can be very different for two separate activity trackers, even if they were worn by the same person during the same time period. An important issue here is that the companies manufacturing the devices consider the underlying algorithms to be their proprietary right, so it is not possible to perform an independent validation of their accuracy, or to perform an independent replication of the findings.

An example of an area that can be of importance for both clinical and personal use is privacy and security issues. It can often be difficult to access the raw data from devices used and users are often not aware of the implications of the terms and conditions of use for the devices. What does the company that offers a particular technological solution do with the personal data, or the objective outcomes that have been recorded? Individuals who consider using technology for self-tracking need to be able to trust that their privacy is fully protected, and that their data are not being used against them.

How can patient-led research and personal science reach their full potential?

"Science and everyday life cannot
and should not be separated."
- Rosalind Franklin

All PhD theses are personal but this one is probably unusually so. I had my first symptoms of symptoms of PD over 35 years ago (my age at onset of PD was 13 years) and around 2010 I decided to start pursuing a PhD. I wanted to complement my lived experience of PD as an illness with the scientific and theoretical knowledge of PD as a disease. I was surprised to realise that the absolute majority of research on selfcare is conducted by persons without lived experience of the disease in question. I therefore decided to make this my field of study and to do so by openly using my own patient experiences as a patient researcher. I am a patient researcher in an academic context and conduct both conventional scientific studies (**Chapters 4 & 5**) as well as contribute to the emerging field of personal science (**Chapters 2 & 3**).

Patient researchers challenge the roles in research

Over time, the roles of patients in research have evolved, see **Chapter 6** and **Figure 9.3**. Previously, patients only contributed to research as a passive subject in studies that were designed and conducted by professional researchers. However, today patients

are contributing at all three levels depicted on the left in **Figure 9.3**. A model often used when studying power dynamics between groups interacting in different ways is "the ladder of citizen participation" formulated by Sherry Arnstein in 1969, see **Figure 9.3** (right) [35]. The rungs of the ladder correspond to different levels of participation, where rungs higher up correspond to higher degrees of participation. The patient role of research participant can be seen as equivalent to the non-participation section of Arnstein's ladder. The research partner role can be seen as encompassing the tokenism (making a symbolic effort to be inclusive) section of the ladder and, in well-developed research projects, extend to the partnership rung. The role of patient researcher corresponds to the citizen power section of Arnstein's ladder and, as this entire thesis has demonstrated, patient researchers can contribute to the advancement of science by combining and contrasting the lived and learned experiences.

There is no doubt that personal science and patient researchers can challenge existing paradigms. Patient researchers are uniquely positioned to potentially contribute to bridging the lived and the learned experiences. Importantly, as the example of patient researchers in long-term COVID mentioned in **Chapter 1** demonstrates, the best results are likely to occur when patient researchers collaborate with experienced clinicians and researchers.

Since patient-led research is a relatively new field, specific issues can come to light, one of which is ethical considerations. For patient researchers using their patient experiences in a conventional academic context, usual ethical guidelines and regulations apply. For patient researchers working in the field of personal science however, new challenges with regards to ethical considerations emerge, which are discussed in **Chapter 7**.

Potential of personal science

Personal science has the potential to contribute to selfcare, healthcare, and health. These practices need to be implemented in an appropriate manner. This thesis suggests (**Chapter 4 & 5**) that persons with PD in Sweden have the autonomy and abilities needed for personal science as well as the attitudes and interests necessary to benefit. Although this of course needs to be studied formally, I suspect that persons with PD in other similar countries are not markedly different. However, the desire and certainly the ability to participate in science may very well be different for other cultures, for example those where the role of women in society is less well established than in a country such as Sweden. Ultimately, I would hope that persons living with PD across the entire world, regardless of their gender, ethnicity, or economical background, would be able to benefit from personal science, should they desire to engage in this. Much more work remains in this field to realise that dream.

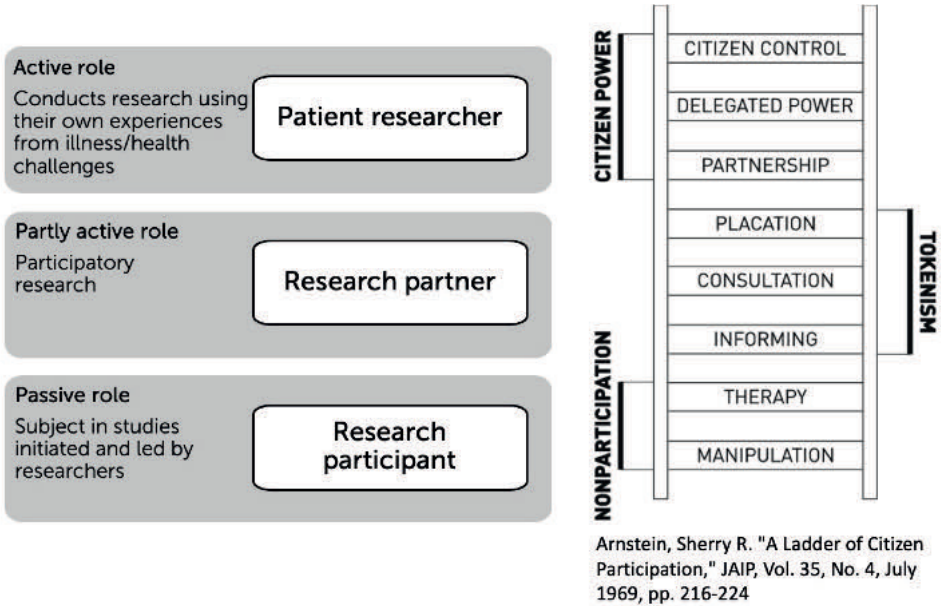


FIGURE 9.3: Patients' roles in research (left) compared to Arnsteins ladder of participation [35] (right).

Personal science is likely to become increasingly important, possibly especially in the context of PD. With the growing evidence that PD is not an homogeneous condition with one single pathophysiology, but, as mentioned in **Chapter 7**, it may be more appropriate to speak of 7 million different types of parkinsonism, namely as many as there are individuals living with this condition in the world [1]. This may mean that, to phrase it extremely, we will need just as many personalised treatment approaches. Personal science is one tool that can bring this approach a step closer to reality.

Furthermore, it would be very unfortunate if personal science were to increase inequalities in health. In future work, aspects of diversity, inclusivity, and equitability will therefore be important, so that factors such as nationality, culture, gender, education levels, and income levels are considered. This thesis indicates that there are important differences in the uptake of practices relating to personal science among persons with PD.

To enable personal science to improve selfcare for PD, structures are needed to learn from the insights made by personal scientists. One way of doing that could be to support groups of persons with PD to conduct personal science projects together, with inspiration taken from research conducted in other fields [18,36,37]. Future efforts should also include structures for disseminating findings (positive and negative) from personal science projects.

Collaborations with healthcare providers

"I don't want to engage with the health system,
I want to engage with life"
- Hugo Campos, Data liberation activist, California

When personal science is done in relation to healthcare, further challenges emerge. For self-recorded data (collected using technology or pen and paper) to be used optimally in the healthcare encounter, data need to be appropriately integrated into the healthcare process. I show in **Chapter 5** that it is not uncommon for persons with PD to show their results from personal science efforts to their healthcare providers. It is, however, less common for healthcare providers to be interested in these data from persons with PD. This is understandable, seeing as healthcare providers rarely have the time or support to work together with persons interested in self-tracking. A vision for the future would be that data collected by persons with PD are seen by all parties as making a significant contribution to the overall quality of care, by both patients and physicians, and that these data can be used to inform genuine shared decision making leading to tailored personalised medical decisions. Much work is, however, needed to achieve that future, both relating to identifying ways of making self-collected data readily available to the clinical team, and also, when using technology, at the level of acceptability and reliability of devices and algorithms. Finally, I want to emphasise once again that it is important that issues around privacy and security of self-recordings are adequately addressed in a transparent way to all parties involved.

Clinicians and persons with PD seem to sometimes have somewhat different views on the uses of technology. A focus group study found that persons with PD saw potential in using technology for their own selfcare, including adjustment of medication [38]. Healthcare professionals in the same study expressed concerns about persons with PD adjusting their medication based on tools that had not been provided to them by the clinic. There are several possible explanations for this reluctance among healthcare professionals: for instance, lack of time, and presumably also well-meaning protection of patients. However, there is also an existing power imbalance between healthcare professionals and patients. In general, physicians do not see patients as equal partners [10,39–44]. The reactions of healthcare professionals were not specifically investigated in this thesis, and this should definitely be the focus of further research. But the results in **Chapter 5** indicate that self-tracking by persons with PD can influence the relationships with healthcare providers both positively and negatively. I know from my own experience that a patient can be knowledgeable, well-read, and autonomous, but during the clinical encounter that does not matter, and it can still be difficult to get clinicians to listen.

An increasing number of scientific papers emphasises the fact that the culture within healthcare can lead to patients being conditioned into passivity and submissiveness. One recent example is a study by Sandén and colleagues [45], where they suggest that a process of "patientification" acts to make patients submissive. Furthermore, they give examples of alternative ways for persons living with long-term conditions to co-create their health and care. Another example is a study by Blease and colleagues, where clinicians' scepticism towards giving patients access to clinical notes are seen as examples of "epistemic exclusion" which negatively impacts patient safety [46]. Furthermore, patients' contributions to clinical knowledge are often undervalued [10,47]. I want to emphasise that I am convinced that this existing culture is not something that healthcare professionals consciously strive to maintain. It is probably more of a somewhat inevitable consequence of the historical difference in the starting point of the relationship between providers and patients; the latter are dependent and traditionally lack knowledge (resulting in uncertainty, anxiety and dependency), whereas the former proactively take a helping and perhaps even paternalistic role, with the best intention to support people and "fix" their problems. It will take a lot of work and considerable effort to create and sustain the more collaborative healthcare culture that we all need and deserve. I am convinced that personal science can assist as a framework in this transformation.

FUTURE DIRECTIONS AND RECOMMENDATIONS

Personal science as an academic field is young, the first use in a scientific publication was in 2016 [4]. The terms and concepts in the field are not always unambiguously defined and misconceptions are not uncommon. One main challenge is therefore to continue developing the theoretical understanding and practices of personal science.

Recommendations for academics and clinicians

In **Table 9.1**, I have summarised my recommendations for future directions to optimise personal science and patient-led research in PD.

TABLE 9.1: Recommendations – How to optimise personal science and patient-led research in PD.

Recommendations – How to optimise personal science and patient-led research in PD
Develop theoretical understanding and personal science methods & tools
• Increase our understanding of the burden of self-tracking • Further explore the use of digital technologies in personal science for PD (including issues relating to data ownership/accessibility, interoperability, and privacy/security) • Develop and test joint group research in PD • Understand and increase acceptance and support by clinicians • Develop structures for disseminating results (positive and negative) of personal science projects, including ascertaining a seamless integration into the primary healthcare process
Ethical issues
• Diversity, inclusivity, and equitability o Conduct personal science and patient-led research in other contexts (different countries, genders, cultures, ethnicities, education levels, income levels, etc.) • Develop ethical frameworks specifically for personal science o Appropriate peer-review process for studies o Protect personal scientists against potential self-harm
Target populations
• for which persons with PD is personal science suitable/not suitable? • for which persons with PD is patient-led research suitable/not suitable?
Personal science education
• for persons with PD o how to use it for your own selfcare o how to use it to also help others with PD o patient researcher: how to also contribute to developing the research field of personal science (scientific training) • for healthcare professionals o identify in which ways personally collected data could be integrated into the primary healthcare process, for example coupled to the electronic health record o how to support persons with PD to use personal science for their selfcare (individually or in groups) o how to work with and support patient researchers and develop the research field of personal science together with them • for researchers o provide a further evidence base for the merits of personal science o how to work with personal science data o how to work with and support patient researchers and develop the research field of personal science together with them
Further development of patient researchers
• structures for formally recognising contributions of patient researchers • encouraging and supporting patients to take the lead in research projects • educate colleagues without lived experience on contributions of patient researchers

9

Living with a complex chronic disease, like PD, affects your whole existence. It affects your future: "How fast will my PD progress?" "Will I be able to babysit my potential future grandchildren without worrying about falling?" It affects your past: "Could I have done anything differently to avoid developing PD?" And it certainly affects your present.

I have done a lot of work over the years, learning as much as I could about PD, about its symptoms and signs, about medications and non-pharmacological treatments, and about different kinds of research. During a visit with my neurologist a couple of years ago, he even told me that I know more about PD research than he does.

I have certainly spent a lot of time learning about PD. And it has paid off, I wouldn't do as well as I am if I hadn't done all that work. Because no-one can understand my PD and what I need as well as I can!

Recommendations for persons with PD

By nature, a PhD thesis is mainly written for an academic readership. Being a person with PD myself, I also want to address persons with PD reading my thesis. I therefore want to end by listing some implications of my work for other persons with PD as well as a few recommendations. The advice I give is a mix of take-home messages from the scientific literature I have read, my own experiences, and the academic work that I present in this thesis.

Build your support network

When you live with a complex chronic condition, like PD, you need to have a good support team, you need to work with good people. First, you have to accept your own responsibility as the "team leader". You are the only one with complete access to the full picture of your life, priorities, sensations, and experiences. Below, I present what I see as the most important building blocks of a personal support team for persons with PD.

Your closest family and friends

Your closest family and friends are your first line of support. They can help you with practical tasks as well as being your "external mirror". In my experience our closest family and friends can be very helpful when reflecting on potential new symptoms or emerging concerns.

Your medical team

PD is best managed clinically in a multidisciplinary team setting [1,48]. A wonderful way to illustrate the relationship between persons with PD and their medical team can be seen in **Figure 9.4**, which is reproduced (with permission) from an article by Bloem, Okun, and Klein [1]. In the Figure, persons with PD are placed in the centre, as the sun in their own universe. Around them, different members of their medical team are placed in circles, closer to the centre for more important members.

As reflected by the position on the innermost circle in **Figure 9.4**, your medical team needs to include a neurologist (or in some settings, a geriatrician). Ideally, this would be someone with dedicated experience in movement disorders, or a clinician with similar such knowledge. It is important that you find a medical specialist that you can trust and who can meet you and your needs where you are at that time. Depending on the country where you live, and also depending on the severity or complexity of your PD, it is likely that you will see this person only once or twice a year (note that even in a well-developed countries such as the United States, about one third of patients has virtually no access to a neurologist). The difficulty in getting access to a medical specialist, and the relatively low frequency of these contacts, both make it even more important to establish a fruitful collaboration grounded in mutual respect for the other person's competences and expertise.

Also as seen in the circle closest to the centre, you will need to have a more general physician on your team as well, to help you with other health concerns. In some countries this is known as a general practitioner or family practitioner.

In the second circle from the centre in **Figure 9.4**, several other important team members can be found. One example is a person that can help you with your physical activity and exercise. Preferably that person would be a physical therapist specialising in PD but unfortunately not all countries or health systems provide that competence. As an alternative, other professionals with deep knowledge of the physical manifestations of the human body can provide value. It is likely and probably also desirable that you see this person more often than your neurologist.

I want to point out that, unfortunately, the depiction in **Figure 9.4** is not yet the reality for many if not most persons with PD, not even in well-developed countries with a good healthcare system such as Sweden or the Netherlands. Ideally, your personal needs would govern which additional competencies from circles two and three that you would get access to, for example the parkinson's nurse, speech and language pathologist, occupational therapist, and psychologist. Depending on what is available in your particular health system or country, your medical team may include medical professionals from these categories.

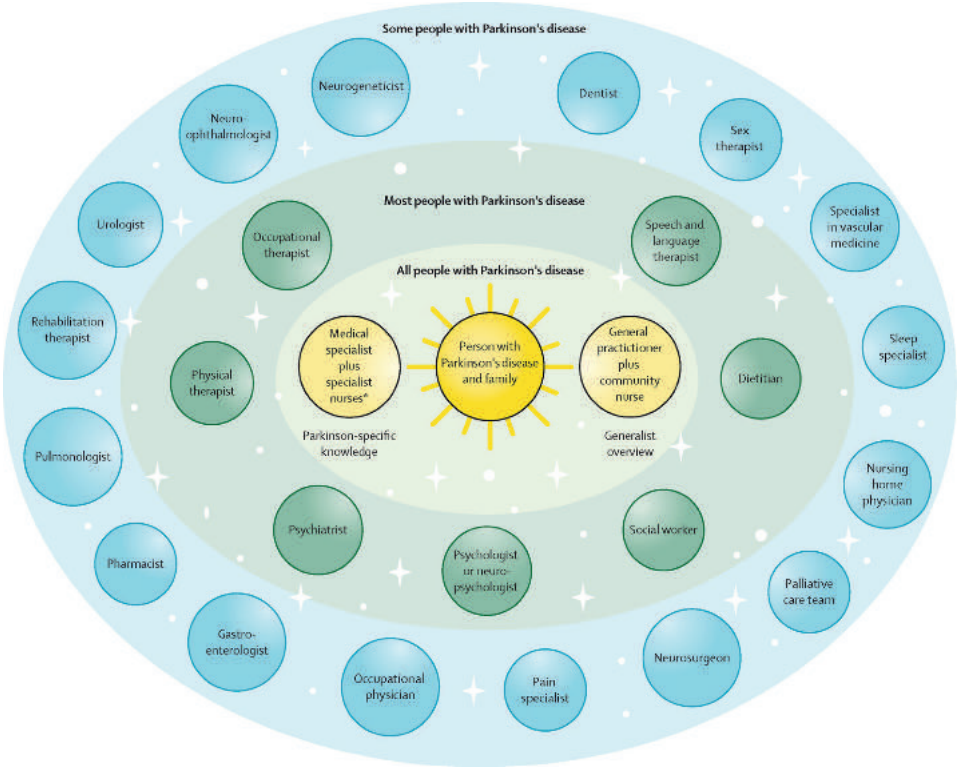


FIGURE 9.4: Professional disciplines involved in the multidisciplinary care for people with Parkinson's disease. Figure [1], reprinted from The Lancet, Vol. 397, Issue 10291, Bastiaan R Bloem, Michael S Okun, Christine Klein, "Parkinson's disease", Pages No. 2284-2303, Copyright (2021), with permission from Elsevier.

Other persons with PD

I have learned so much from other persons with PD. In fact, I have probably learned more about PD from other persons with PD than I have learned from my healthcare providers. What I have learned from my healthcare providers has been an invaluable base for me to start with but frankly, very few healthcare providers genuinely know what it is like to live with PD so their contribution can only go so far.

There are many ways to connect with other persons with PD: patient organisations, in-person support groups, conferences, and, of course, a wide range of online options to connect. In my opinion, many of them are good and serve slightly different purposes. You just have to try to find your own best fit. It is also important to emphasise that not every individual living with PD automatically sees the merits of meeting fellow persons with PD. You might be afraid of being confronted with your own future, for example when meeting persons in a more severe disease state. I was myself hesitant to meet

other persons with PD when I was early in my PD journey. It is important to make sure that it is easy to find other persons with PD who are in a comparable phase of their life, and in a comparable phase of their PD. One possibility is web-based communities [49–52].

Educate yourself

Knowledge and learning are key to successfully managing a complex chronic condition, like PD. Research from different fields has demonstrated that the culture within healthcare can lead to patients being conditioned into being passive and unnecessarily dependant on healthcare providers. In this thesis (**Chapter 4**) I demonstrate that knowledge about PD can lead to increased autonomy, and it can also enable you to engage in discussions with your medical team at a higher level. It is important to remember that healthcare providers' view on selfcare is mostly restricted to the clinical context, while people living with chronic and long-term conditions see selfcare as a more holistic concept, and as a means to achieve individual health.

This means that patient education efforts offered by healthcare may have a similar restricted view of selfcare and what we need to be able to successfully manage PD. Patient education offered by healthcare can be a good start but in addition, you should also build your own understanding of your own individual situation.

Track purposefully and moderately

This whole thesis is about how personal science can be used to improve selfcare for PD and I am convinced that personal science and self-tracking, if used optimally, can provide a lot of benefit to persons with PD. Here, optimally means to make sure that the balance between benefits and burdens of tracking tips towards benefits. More discussion on that topic can be found in the sections *Using personal science to improve selfcare* and *Benefits and burdens* above.

The benefits of tracking can be leveraged by tracking purposefully and moderately. By that I mean that tracking should be used for specific purposes and only for limited periods of time. It can for example mean that you track episodically with an acceptable frequency, probably throughout the entire course of your PD, since each new phase comes with its new challenges. For your tracking to be able to contribute to your own learning, it is crucial that you define questions that are important to you and identify what data you would need to collect to answer those questions. Technology can in some cases facilitate the data collection, but it is not always necessary, especially for conditions currently lacking an objective biomarker, such as PD. Pen and paper can be surprisingly powerful tracking tools. For personal science, the context in which the data are collected can say a lot about the phenomenon you are tracking, and it

is therefore important to also capture that, which can often be easier to do with pen and paper.

It is important to remember that specific interventions, such as combinations and doses of medications, are often not generalisable. However, the methods used for deciding if for example a specific medication or dose works or not, are often generalisable. This means that there is a lot to be learned from other persons with PD when it comes to the methods they use for tracking.

When the data are collected, it is time for the really interesting work, namely to start reasoning with your data and to make sense of what this means to you and to the care or other support that you should receive. This is where you use your data (in the form of numbers or simply observations and/or notes) to reason and think about what they mean to you in your context. The magic doesn't happen during tracking, but only as soon as you are reasoning with your self-collected data and associated experiences.
Lastly, I want to emphasise that, in my experience, tracking for the sake of tracking is rarely worth the effort. By this I mean that for the effort of tracking to be worth it, you should have an idea of what you want to use it for. I have also found it to be important to make sure that what you decide to track is actionable.

I hope that this thesis can provide some new ideas for you to explore. I also hope that we, the community of persons living with PD, together with healthcare professionals, scientists and other people around us, jointly can develop new ways to keep living a good life, with PD.

Useful websites

I want to end by giving recommendations of some useful websites. Of course, I am aware that websites are not always constant. I have therefore attempted to identify sites that I expect to remain online for a reasonable time. This list is not intended to be a comprehensive overview but should be seen as offering some illustrative examples.

International

National Library of Medicine, PubMed: https://pubmed.ncbi.nlm.nih.gov
World Parkinson Coalition: https://www.worldpdcoalition.org
World Parkinson Congress 2023: https://wpc2023.org
Parkinson & Movement Disorder Alliance: https://www.pmdalliance.org
Rock Steady Boxing International: https://rocksteadyboxing.org

Swedish

1177 Vårdguiden: https://www.1177.se/
Mina vårdkontakter och journal: https://journalen.1177.se/
ParkinsonFörbundet: http://www.parkinsonforbundet.se
Neuro: https://neuro.se

Dutch

ParkinsonNL: https://www.parkinson.nl
ParkinsonNet: https://www.parkinsonnet.nl
ParkinsonNext: https://www.parkinsonnext.nl
Punt voor Parkinson: https://puntvoorparkinson.nl
Parkinson Vereniging: https://www.parkinson-vereniging.nl
ParkinsonSport NL: http://www.parkinsonsport.nl
Rock Steady Boxing in Nederland: http://rocksteadyboxing.nl

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APPENDICES

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Dear Bas, I don't think that you can understand how much it means to me to be able to finish my thesis with you as my main supervisor. I remember the first time we met, in Nijmegen in May 2011, at the conference that was organised in connection with a PhD defence. There and then, I decided that some day in the future, I was going to work with you. And through a series of serendipitous events, that wish did come true!
Dear Maria, it's no exaggeration to say that we've been through a lot together... Maybe if you'd known about the challenges in advance, you wouldn't have said yes to supervising me? (Don't answer that! ☺) But seriously, I can't think of anyone else that I would rather have wanted to experience this journey with than you! I have learned so much from you, about scientific reasoning, academic writing, how to be a great teacher, about UNESCO's World Heritage sites and so much more. Now it's finally time to celebrate!
Dear Martijn, ever since we first met at the QSEU conference in 2011 I have benefitted from your support. You are always smiling, always have a kind word to say, and are always ready to help. Your feedback and questions are always on the exact right level to make me think about important issues one more time and realise what was missing. I am so very happy to have finished my PhD with you!

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This thesis also makes me a little sad, because some of the people I want to thank are no longer with us. I hope that I was able to express to them how important they were to me and my work while they were alive: Dan Grandér, Tom Isaacs, Lars-Olof Lanebo, Duncan Neuhauser, Michael Seres, Rosamund Snow, and my father Thomas Södergren, who passed away in 1990.

And, to my biggest fans and harshest critics, my reasons for doing all the hard work: Per and Isaac: Thank you! Your support makes me a better person, I love you to the moon and back!

ABOUT THE AUTHOR

Sara Riggare was born on February 19th 1971 in Solna, Sweden. Around 1984, she started noticing the first symptoms of what would later be diagnosed as Parkinson's disease. Sara graduated with a Master's in Engineering from The Royal Institute of Technology in Stockholm in 1994. Thereafter she worked with environmental risk assessments and scenario analyses until she in 2010 decided that she wanted to combine her engineering skills with her patient experiences to try to improve the situation for herself and others with chronic diseases.

Sara started her PhD training in 2012 at Karolinska Institutet in Stockholm, Sweden, under the supervision of Prof. Dr. Staffan Lindblad. Ass. Prof. Dr. Maria Hägglund took over as main supervisor in 2014. In 2020, Sara's PhD project was transferred to Radboud University Medical Center in Nijmegen, The Netherlands where she worked under the supervision of Prof. Dr. Bas Bloem (Radboudumc Center of Expertise for Parkinson & Movement Disorders) and Dr. Martijn de Groot (Radboudumc Health Innovation Labs). Sara's research, which is presented in this thesis, is focused on exploring methods for how individuals living with health challenges (also known as patients) can use their own observations to find answers to their personal questions and to improve their condition. She is working with Ass. Prof. Dr. Maria Hägglund at the Medtech Science & Innovation Centre, Uppsala University, Sweden.

Sara is married to Per, they live in Stockholm, Sweden, and are parents to Isaac (born 2003).

LIST OF PUBLICATIONS

Published articles in this thesis:

1. **Riggare S**, Hägglund M. Precision Medicine in Parkinson's Disease - Exploring Patient-Initiated Self-Tracking. J Parkinsons Dis. 2018;8(3):441-446. doi: 10.3233/JPD-181314. PMID: 30124453
2. **Riggare S**, Unruh KT, Sturr J, Domingos J, Stamford JA, Svenningsson P, Hägglund M. Patient-driven N-of-1 in Parkinson's Disease. Lessons Learned from a Placebo-controlled Study of the Effect of Nicotine on Dyskinesia. Methods Inf Med. 2017 Oct 24;56(99):e123-e128. doi: 10.3414/ME16-02-0040. Epub 2017 Oct 24. PMID: 29064509
3. **Riggare S**, Höglund PJ, Hvitfeldt Forsberg H, Eftimovska E, Svenningsson P, Hägglund M. Patients are doing it for themselves: A survey on disease-specific knowledge acquisition among people with Parkinson's disease in Sweden. Health Informatics J. 2019 Mar;25(1):91-105. doi: 10.1177/1460458217704248. Epub 2017 Apr 23. PMID: 28434277
4. **Riggare S**, Scott Duncan T, Hvitfeldt H, Hägglund M. "You have to know why you're doing this": a mixed methods study of the benefits and burdens of self-tracking in Parkinson's disease. BMC Med Inform Decis Mak. 2019 Aug 30;19(1):175. doi: 10.1186/s12911-019-0896-7. PMID: 31470832
5. **Riggare S.** Patient researchers - the missing link? Nat Med. 2020 Oct;26(10):1507. doi: 10.1038/s41591-020-1080-4. PMID: 33029015
6. **Riggare S**, Hägglund M, Bredenoord AL, de Groot M, Bloem BR. Ethical Aspects of Personal Science for Persons with Parkinson's Disease: What Happens When Self-Tracking Goes from Selfcare to Publication? J Parkinsons Dis. 2021;11(4):1927-1933. doi: 10.3233/JPD-212647. PMID: 34120915

Other articles:

7. **Riggare S**, Stamford J, Hägglund M. A Long Way to Go: Patient Perspectives on Digital Health for Parkinson's Disease. J Parkinsons Dis. 2021;11(s1):S5-S10. doi: 10.3233/JPD-202408.PMID: 33682728
8. Salmi L, Brudnicki S, Isono M, **Riggare S**, Rodriquez C, Schaper LK, Walker J, Delbanco T. Six countries, six individuals: resourceful patients navigating medical records in Australia, Canada, Chile, Japan, Sweden and the USA. BMJ Open. 2020 Sep 15;10(9):e037016. doi: 10.1136/bmjopen-2020-037016. PMID: 32933961
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10. Domingos J, Radder D, **Riggare S**, Godinho C, Dean J, Graziano M, de Vries NM, Ferreira J, Bloem BR. Implementation of a Community-Based Exercise Program for Parkinson Patients: Using Boxing as an Example. J Parkinsons Dis. 2019;9(3):615-623. doi: 10.3233/JPD-191616.PMID: 31282426
11. **Riggare S.** E-patients hold key to the future of healthcare. BMJ. 2018 Feb 26;360:k846. doi: 10.1136/bmj.k846.PMID: 29483151
12. **Riggare S**, Unruh KT. Patients organise and train doctors to provide better care. BMJ. 2015 Nov 30;351:h6318. doi: 10.1136/bmj.h6318.PMID: 26620969

PORTFOLIO

PhD candidate:	Sara Viktoria Södergren Riggare
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	March 2012 - December 2019
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	January 2020 - March 2022
Research school:	Radboud University Nijmegen
	Donders Institute for Brain Cognition and Behaviour
Department:	Radboudumc Neurology Department
	Donders Centre for Cognitive Neuroimaging
Supervisor:	Prof. Dr. Bas Bloem
Co-Supervisors:	Dr. Martijn de Groot, Ass. Prof. Dr. Maria Hägglund

PhD training

Subject	Location	Year
General courses and seminars		
Public Health Intervention and Implementation Research	Karolinska Institutet	2012
To Communicate Science in Different Contexts	Karolinska Institutet	2013
Basic Course in Medical Statistics	Karolinska Institutet	2013
Philosophy of Science and Research Ethics	Karolinska Institutet	2013
Write your Research Results and get Them Published	Karolinska Institutet	2014
Methods for Qualitative Content Analysis	Karolinska Institutet	2017
PhD Introduction	Donders Institute	2020
Donders Graduate School	Donders Institute	2020-2021
Scientific Integrity Course	Donders Institute	2021
Conferences		
3rd World Parkinson Congress	Montreal, Canada	2013
Medicine 2.0 World Congress	London, UK	2013
4th World Parkinson Congress	Portland, USA	2016
International Forum for Quality and Safety in Healthcare	London, UK	2017
International Forum for Quality and Safety in Healthcare	Amsterdam, NL	2018
International Forum for Quality and Safety in Healthcare	Glasgow, UK	2019
Teaching		
Several teaching courses and seminars in health informatics, movement disorders, patient safety, and digital health at Karolinska Institutet and Uppsala University, Sweden. Supervised master thesis work for students in health informatics, Karolinska Institutet.		2013 - 2021
Other activities		
Member of the BMJ patient panel		2014 -
Member of the steering committee for the World Parkinson Congress		2019 -
Member of the editorial board for the journal Digital Biomarkers		2019 -

RESEARCH DATA MANAGEMENT

This thesis is based on the results of human studies, which were conducted in accordance with the principles of the Declaration of Helsinki. All research data for this thesis were collected at Karolinska Institutet in Stockholm, Sweden.

All participants were informed about the study in question and how their personal data would be handled and anonymously stored. In the studies in **Chapters 2 and 3**, the PhD candidate was both the researcher and the person studied. The research in these studies was not subjected to an ethical review board. Reflections and perspectives on ethics for self-research are shared in **Chapter 7**. The Regional Ethical Review Board in Stockholm, Sweden gave a positive advice for the studies in **Chapters 4 and 5** according to decision 2015/1572-31/4. Participants are not named or identified in publications.

Data for the surveys in **Chapters 4 and 5** were collected by means of online surveys. The surveys were each online for a period of four weeks, after which the data were downloaded to a secure location and the online versions were deleted. The survey data were analysed using Microsoft Excel and a statistical online service (http://quantpsy.org).

All interview participants (**Chapter 5**) provided written consent. The interviews were audio recorded and stored marked with a code, related to time, location, interviewer, and interviewee. The audio files are stored securely in a digital format and will be deleted after 10 years. No personal data are stored with the audio files and only researchers directly involved with the project are given access. The code key for connecting the audio files with the correct code is stored separately from the audio files. The interviews have been transcribed without including personal identifiers.
Data at Karolinska Institutet are stored according to applicable rules and guidelines, which means that research data are saved for at least 10 years.

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, 17 June 2008
- Maaike Bakker. Supraspinal control of walking: lessons from motor imagery. Radboud University Nijmegen, 27 May 2009
- W. Farid Abdo. Parkinsonism: possible solutions to a diagnostic challenge. Radboud University Nijmegen, 7 October 2009
- Samyra H.J. Keus. Physiotherapy in Parkinson's disease. Towards evidence-based practice. Leiden University, 29 April 2010
- Lars B. Oude Nijhuis. Modulation of human balance reactions. Radboud University Nijmegen, 29 November 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, 29 November 2010
- Rick C.G. Helmich. Cerebral reorganization in Parkinson's disease. Radboud University Nijmegen, 24 May 2011
- Charlotte A. Haaxma. New perspectives on preclinical and early stage Parkinson's disease. Radboud University Nijmegen, 6 December 2011
- Johanna G. Kalf. Drooling and dysphagia in Parkinson's disease. Radboud University Nijmegen, 22 December 2011
- Anke H. Snijders. Tackling freezing of gait in Parkinson's disease. Radboud University Nijmegen, 4 June 2012
- Bart F.L. van Nuenen. Cerebral reorganization in premotor parkinsonism. Radboud University Nijmegen, 22 November 2012
- Wandana Nanhoe-Mahabier. Freezing of physical activity in Parkinson's disease, the challenge to change behavior. Radboud University Nijmegen, 13 February 2013
- Marlies van Nimwegen. Promotion of physical activity in Parkinson's disease, the challenge to change behavior. Radboud University Nijmegen, 6 March 2013
- Arlène D. Speelman. Promotion of physical activity in Parkinson's disease, feasibility and effectiveness. Radboud University Nijmegen, 6 March 2013
- Tjitske Boonstra. The contribution of each leg to bipedal balance control. University Twente, 6 June 2013
- Marjolein A van der Marck. The Many faces of Parkinson's disease: towards a multifaceted approach? Radboud University Nijmegen, 10 January 2014
- Katrijn Smulders. Cognitive control of gait and balance in patients with chronic stroke and Parkinson's disease. Radboud University Nijmegen, 21 May 2014
- Marjolein B. Aerts. Improving diagnostic accuracy in parkinsonism. Radboud

University Nijmegen, 27 June 2014

- Maartje Louter. Sleep in Parkinson's disease. A focus on nocturnal movements. Radboud University Nijmegen, 13 February 2015
- Frederick Anton Meijer. Clinical Application of Brain MRI in Parkinsonism: From Basic to Advanced Imaging, Radboud University Nijmegen, 23 June 2015
- Jorik Nonnekes. Balance and gait in neurodegenerative disease: what startle tells us about motor control, Radboud University Nijmegen, 2 September 2015
- Martijn van der Eijk. Patient-centered care in Parkinson's disease. Radboud University Nijmegen, 1 December 2015
- Ingrid Sturkenboom. Occupational therapy for people with Parkinson's disease: towards evidence-informed care. Radboud University Nijmegen, 11 February 2016
- Merel M. van Gilst. Sleep benefit in Parkinson's disease. Radboud University Nijmegen, 13 April 2016
- Arno M. Janssen. Transcranial magnetic stimulation - measuring and modeling in health and disease. Radboud University Nijmegen, 2 June 2016
- Annette Plouvier. De ziekte van Parkinson, een gezamenlijke reis van huisarts en patient. Radboud University Nijmegen, 15 June 2017
- Nico Weerkamp. Parkinson's disease in long-term-care facilities. Radboud University Nijmegen, 1 September 2017
- Digna de Kam. Postural instability in people with chronic stroke and Parkinson's disease: dynamic perspectives. Radboud University Nijmegen, 4 October 2017
- Freek Nieuwhof. The complexity of walking: Cognitive control of gait in aging and Parkinson's disease. Radboud University Nijmegen, 27 October 2017
- Koen Klemann. A molecular window into Parkinson's disease. Radboud University Nijmegen, 3 November 2017.
- Claudia A. Barthel. Moving beyond freezing of gait in Parkinson's disease. Radboud University Nijmegen, 3 April 2018.
- Esther Bekkers. Freezing and postural control in Parkinson's disease. Defense at KU Leuven, 15 May 2018.
- Erik te Woerd. Feeling the beat: The neurophysiology of cueing in Parkinson's disease. Radboud University Nijmegen, 18 January 2019.
- Ana L. Silva de Lima. Quantifying Parkinson's disease: the use of technology for objective assessment of motor symptoms. Radboud University Nijmegen, 26 March 2019.
- Nicolien M. van der Kolk. Towards a prescription for exercise for persons with Parkinson's disease. Radboud University Nijmegen, 30 October 2020.
- Michiel Dirkx. Neural mechanisms of Parkinson's tremor. Radboud University Nijmegen, 2 November 2020.
- Anna Santaella. Tackling Parkinson's disease: a proteomic approach to biomarkers and regenerative therapy. Radboud University Nijmegen, 22 November 2020.

- Sjors C.F. van de Weijer. Digital-technology enabled home health care: gamification in online cognitive therapies for Parkinson's disease. Maastricht University, 1 September 2021.
- Taina Marques. Discriminating parkinsonian disorders: using fluid biomarkers to improve early diagnosis. Radboud University Nijmegen, 9 September 2021.
- A.L.A.J. Hommel. Impairment and disability in late-stage parkinsonism. Radboud University, 27 September 2021.
- Floris Vlaanderen. Towards seamless and sustainable care for Parkinson's disease. Radboud University Nijmegen, 27 October 2021.

Non-Parkinsonian disorders of movement

- Sascha Vermeer. Clinical and genetic characterization of autosomal recessive cerebellar ataxias. Radboud University Nijmegen, 5 April 2012
- Susanne T. de Bot. Hereditary spastic paraplegias in the Netherlands. Radboud University Nijmegen, 20 December 2013
- Catherine C.S. Delnooz. Unraveling primary focal dystonia. A treatment update and new pathophysiological insights. Radboud University Nijmegen, 7 January 2014
- Ella MR Fonteyn. Falls, physiotherapy, and training in patients with degenerative ataxias, 29 June 2016
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- Gerald JD Hengstman. Myositis specific autoantibodies, specificity and clinical applications. Radboud University Nijmegen, 21 September 2005
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- Bert de Swart. Speech therapy in patients with neuromuscular disorders and Parkinson's disease. Diagnosis and treatment of dysarthria and dysphagia. Radboud University Nijmegen, 24 march 2006
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- Mijke M.M. Verhagen. Ataxia-Telangiectasia from childhood to adulthood. Radboud University Nijmegen, 8 October 2012
- Ilse Arts. Muscle ultrasonography in ALS. Radboud University Nijmegen, 31 October

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The DGCN tracks the career of PhD graduates carefully. More than 50% of PhD alumni show a continuation in academia with postdoc positions at top institutes worldwide, e.g. Stanford University, University of Oxford, University of Cambridge, UCL London, MPI Leipzig, Hanyang University in South Korea, NTNU Norway, University of Illinois, North Western University, Northeastern University in Boston, ETH Zürich, University of Vienna etc.. Positions outside academia spread among the following sectors: specialists in a medical environment, mainly in genetics, geriatrics, psychiatry and neurology. Specialists in a psychological environment, e.g. as specialist in neuropsychology, psychological diagnostics or therapy. Positions in higher education as coordinators or lecturers. A smaller percentage enters business as research consultants, analysts or head of research and development. Fewer graduates stay in a research environment as lab coordinators, technical support or policy advisors. Upcoming possibilities are positions in the IT sector and management position in pharmaceutical industry. In general, the PhDs graduates almost invariably continue with high-quality positions that play an important role in our knowledge economy.

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