

**Occupational therapy for people  
with Parkinson's disease:**  
towards evidence-informed care



Ingrid Sturkenboom

# Occupational therapy for people with Parkinson's disease: towards evidence-informed care

**Occupational therapy for people with Parkinson's disease:** towards evidence-informed care, PhD Thesis, Radboud University Nijmegen, The Netherlands, 2016

The work presented in this thesis was carried out within the Radboud Institute for Health Sciences, at the department of Rehabilitation, Radboud university medical center.

Financial support for the feasibility study was provided by a grant from Fonds Nuts Ohra and for the main studies by a combined grant from Prinses Beatrix Spierfonds and Parkinson Vereniging.

Printing and dissemination of this thesis was financially supported by the Radboud university medical center, Prinses Beatrix Spierfonds, Parkinson Vereniging and Stichting Parkinson Nederland.

## Cover concept

Juan Carlos Gonzalez Pelayo, adapted design of the Parkinson Tulip

The tulip on the cover has been the international symbol for people with Parkinson's disease since 2005. In 1980 a Dutch bulb farmer with Parkinson's disease cultivated the Dr James Parkinson tulip. An American painter, Karen Painter, who also had Parkinson's disease, designed the drawing in which the leaves form the letters P and D (Parkinson's disease). The symbol was used on the cover of the Dutch guidelines of Occupational Therapy, which is the basis of the intervention evaluated in this thesis. The O and T have now been added symbolising the (intended) enabling/empowering role of occupational therapy for people with Parkinson's.

## ISBN

978-94-6259-989-5

## Design/layout

Promotie In Zicht, Arnhem

## Print

Ipskamp Drukkers, Enschede

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## Proefschrift

Ter verkrijging van de graad van doctor  
aan de Radboud Universiteit Nijmegen  
op gezag van de rector magnificus,  
volgens besluit van het college van decanen  
in het openbaar te verdedigen op donderdag 11 februari 2016  
om 12.30 uur precies

door

**Ingrid Helena Wilhelmina Maria Sturkenboom**

geboren op 5 oktober 1969

te Houten

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“Such understated power here  
in these tottering dancers  
who exert stupendous effort  
on tasks most view as insignificant.

Such quiet beauty here,  
in these soft voiced, stiff limbed people;  
such resolve masked by each placid face.

There is immensity required  
in growing small,  
so bent on such unbending grace.”

*Robin Morgan, taken from her poem*

*‘no signs of struggle’*

*at TED Women 2015, USA*

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## Chapter 1

### **General introduction\* and outline of the thesis**



\* Parts of the text are based on a book chapter: Sturkenboom IH, Keus SH, Munneke M; Bloem BR: Physical and occupational therapy. In: *Handbook of Parkinson's Disease* (2013)

People living with Parkinson's disease (PD) and their families may come across many challenges and barriers in their daily lives. The mounting disease symptoms and subsequent changes in daily activities and personal identity require continuous creativity and flexibility to adapt. Managing change becomes a prominent part of life.<sup>2</sup> According to a proposed new concept of health, namely 'the ability to adapt and self-manage',<sup>3</sup> this process can be seen as a continuous effort to achieve optimal health (i.e. a good quality of life). Many professionals may offer support to persons with PD and their families to minimise disability and to help them deal better with the impact of PD. Occupational therapy is one of these professional disciplines and focuses on enabling persons to adapt and self-manage the challenges they encounter in meaningful activities and roles.

The main aim of this thesis is to describe the systematic evaluation of a home-based occupational therapy intervention for people living with PD and their primary caregivers. First, we explored the feasibility of the intervention and a trial. Subsequently, we assessed efficacy and cost-effectiveness of the intervention. We concluded with a process evaluation of treatment delivery and mediating factors.

This chapter provides an overview of the consequences of PD, the role of occupational therapy within a multidisciplinary team approach to Parkinson care, and offers considerations for outcome measurement. The chapter concludes with an outline of this thesis, which follows the different phases of the Medical Research Council's Framework for developing and evaluating complex interventions.<sup>4,5</sup>

## Parkinson's disease

Parkinson's disease is a neurodegenerative disorder that progressively affects the dopaminergic and non-dopaminergic areas of the brain. The exact aetiology for this neuronal degeneration is still unknown and there is no medical cure available. PD is the second most common neurodegenerative disorder after Alzheimer disease. In industrialised countries the estimated prevalence is 0.3% in the general population and 1% in people above 60 years old.<sup>6,7</sup> Considering the ageing population, the absolute number of people with PD is expected to double in the next twenty years.<sup>8</sup>

PD results in a complex presentation of motor and non-motor symptoms. The various symptoms are summarised in Box 1. The clinical diagnosis is based on the (asymmetrical) presence of bradykinesia plus rigidity and/or a resting tremor.<sup>9,10</sup> Postural instability is a cardinal feature in more advanced disease stages.<sup>9,10</sup> Additionally, certain contra indications for the diagnosis PD (so called 'red flags') should be excluded. Examples of red flags are early postural instability and a poor response to dopaminergic treatment. Although the diagnosis can be made when motor features are present, there is likely be a long premotor phase with specific non-motor features.<sup>10,11</sup>

**Box 1** Parkinson disease symptoms

The number and severity of symptoms varies between individuals and increases as the disease progresses. Severity of symptoms may vary throughout the day depending on the response to medication (wearing off, dyskinesias).

**Motor symptoms<sup>10</sup>**

Bradykinesia, 4-6 Hz resting tremor, rigidity and postural instability are the classical motor symptoms of PD. Note that impaired balance early in the disease is a contra-indication for the diagnosis of Parkinson's disease. Other motor symptoms include impaired gait pattern (e.g. shuffling gait, freezing), reduced control of voluntary movements, postural abnormalities, masked face, dysarthria, and dysphagia (e.g. drooling).

**Non-motor symptoms<sup>11</sup>**

Impairments in mental functions may include: deficits in higher level cognitive functions (e.g. executive dysfunction, mental rigidity), dementia, personality change, visuospatial perception disorder, impaired drive (i.e. apathy), depression, anxiety.

Sleep disorders include REM sleep behaviour disorder, insomnia, excessive daytime sleepiness.

Sensory problems include visual dysfunction, proprioceptive dysfunction, impaired smell, pain.

Autonomic symptoms such as constipation, nocturia, impaired sexual function

Fatigue is a common complaint that may be related to both motor and non-motor problems.

Because there is no cure for PD, medical and surgical treatment can only focus on relieving symptoms. This symptomatic treatment is only partially effective and also complicated by dose-limiting side effects. Therefore, persons with PD have to manage the effects of the diagnosis and the complex range of often debilitating symptoms and signs.

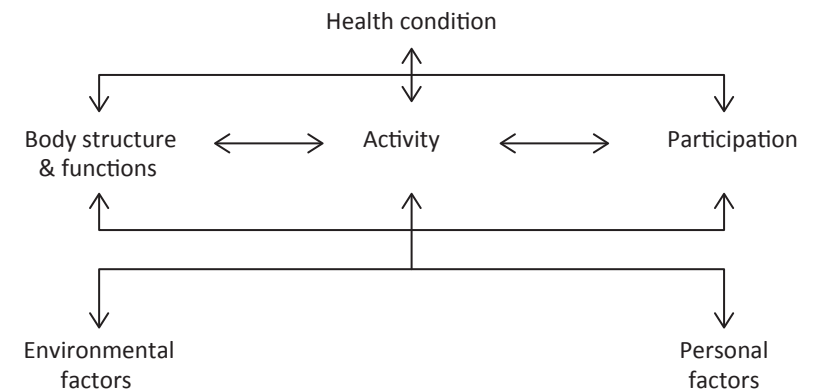
**Impact of Parkinson's disease on the person**

The impact of the disease can be described using the World Health Organization's model of the International Classification of Functioning, Disability, and Health (ICF).<sup>12</sup> The model is illustrated in Figure 1 and exemplifies the dynamic interaction between body structure and functions, activities, participation and contextual factors. This indicates the impact of disease will be different for every individual depending on all the elements in the model. How the person can manage in daily life does not only depend on the type and severity of symptoms, but also personal factors (e.g. coping style, aspirations, age), and physical, social, attitudinal environmental factors.<sup>13</sup> Generally, people with PD experience increasing problems in daily activities and participation as the disease progresses.<sup>14-19</sup>

In depicting the overall impact of disease, health-related quality of life can also be used as a concept. In relation to the ICF, the individual's lived experience of functioning and disability reflects health-related quality of life.<sup>20</sup> In PD, the motor and non-motor impairments and functioning in the psychosocial and daily activities domains were found to be important determinants for health-related quality of life.<sup>21,22</sup>

**Impairment of body functions and structures**

The many symptoms of PD (Box 1) represent impairments of functions. In addition, there may be impairments as a result of side effects of the medication, inactivity, falls, or maladaptive coping. Besides direct or indirect consequences of PD, ageing and co-morbid conditions lead to additional impairments and disease burden.<sup>23</sup>



**Figure 1** International Classification of Functioning, Disability and Health (World Health Organization 2001)<sup>12</sup>

**Limitations in activities**

Difficulties in daily activities, without loss of independent function, can be present in early stages of PD.<sup>16, 24, 25</sup> Limitations in basic mobility-related activities like walking, transfers, posture, balance and manual dexterity impact on complex daily activities such as self-care activities, housekeeping, leisure activities, work-related activities or transport. In fact, the limitations are often more apparent in complex daily activities because the attention load and mental flexibility required for these activities further constrains motor performance.<sup>26, 27</sup> If executive impairments are present, the planning and organizing of complex tasks and routines is compromised. This might become evident first in activities like managing medication, preparing a shopping list or administrative tasks.<sup>28</sup> Other non-motor problems that may affect daily activity

performance are visual deficits (i.e. impaired contrast sensitivity) and visuo-spatial difficulties.<sup>29,30</sup> Contextual factors may influence activity restrictions in all diagnostic groups, but in PD there can be very specific environmental constraints. For example narrow spaces, darkness or a crowded environment provoke freezing.<sup>31,32</sup> A stressful context or perceived time pressure aggravates symptoms like tremor and freezing.<sup>33</sup>

### Restrictions in participation

Due to experienced difficulties in activities, people with PD adjust their daily routine and activity patterns. Reported reasons for giving up activities are that the activities can no longer be performed to desired standard, cause embarrassment, require too much time and effort, or are considered too dangerous by those close to them.<sup>34</sup>

Inevitably, PD changes the person's participation pertaining to work, leisure or community and social life.<sup>26,34-38</sup> Employment is often given up early due to the PD's consequences.<sup>36,39-41</sup> In more advanced stages of disease the unpredictability of symptoms creates a feeling of uncertainty and greatly limits the opportunity for spontaneous engagement in (social) activities.<sup>37,42,43</sup> However, studies on participation in PD show there might also be gains. The person may find new valued activities and roles.<sup>35</sup> Better coping with disease is associated with personal characteristics like resilience and optimism.<sup>44,45</sup> Moreover, the importance of adequate social support to maintain participation is highlighted.<sup>43</sup>

### Impact of Parkinson's disease on caregivers

Considering that PD affects the patient and his or her daily life in a multifaceted way, it is not surprising daily lives and wellbeing of partners or other family members involved are also affected.<sup>46-49</sup> Qualitative studies describe how the management of the changes in activities involves the person with PD and the partner as an integrated unit.<sup>2,13</sup> As the disease progresses, the person with PD is likely to be more reliant on assistance that is mainly provided by family members. Healthcare professionals often also expect the caregiver to a 'co-therapist' by assisting in integrating the medication- and rehabilitation regimes in the daily routine of the patient at home.<sup>50</sup> Therefore, the health and well-being of the informal caregivers are an important concern for healthcare and society. There have been a number of studies attempting to elicit what causes the most distress or burden for caregivers of persons with PD, with varying results. In a meta-analysis, de Lau & Au conclude that higher intensity of care, more severe motor symptoms, and higher dependency in activities of daily living (ADL) correlate mostly with caregiver distress.<sup>51</sup>

In the literature it is found that the burden related to psychosocial issues like social isolation, experience of loss, or safety concerns, has higher impact than the physical care burden (e.g. lifting someone).<sup>46,49</sup>

## Multidisciplinary Parkinson's care

To ensure that all factors that influence disease burden can be addressed effectively, an individually tailored and multifaceted approach to care is warranted.<sup>52-55</sup> Moreover, care should attend to the needs of both the person with PD and the families involved.<sup>2</sup> An additional consideration is that the needs of patients and their families change over time as a result of disease progression, the natural course of life and changes in the societal context.

To offer the required multifaceted, person- and family-centred care approach, many professionals from different settings and disciplines may enter the care team. The involvement of a professional may be transient, intermittent or enduring. The dynamic nature of the care team around an individual with PD and his or her family poses challenges for organizing an effective team with optimal collaboration and adequate expertise.<sup>56</sup>

In the past 10 years, a multidisciplinary model and organization of Parkinson care in the Netherlands has evolved, called ParkinsonNet (Figure 2).<sup>57,58</sup> ParkinsonNet is a nationwide network that comprises 66 regional networks of dedicated and trained health professionals in the field of PD in different settings (e.g. community care, hospital care, nursing home care).



**Figure 2** ParkinsonNet regions in the Netherlands; \* regions included in the main trial described in this thesis



The aim of ParkinsonNet is to deliver high quality, individualised and integrated care to all persons with PD and their families. Currently, many different disciplines participate in ParkinsonNet: neurologists, PD nurse specialists, physiotherapists, speech and language therapists, occupational therapists, dieticians, rehabilitation physicians, specialised elderly care physicians, geriatricians, social workers, psychologists and sexologists. Interprofessional collaboration is facilitated in ParkinsonNet through regional network meetings and a web based platform. In our research we aimed to investigate the value of the occupational therapists' role within in the multidisciplinary team.

## Occupational therapy in Parkinson's disease rehabilitation

### The role of occupational therapy in Parkinson's disease rehabilitation

The role of occupational therapy in PD care is enabling the patient to engage in meaningful roles and tasks/activities in the home and community.<sup>1, 53, 59-62</sup> Within the field of occupational therapy this engagement in activities and roles is called *occupational performance*. Enabling occupational performance includes enhancing actual activity performance related to independence and safety or amount of effort and time, and optimising the experience of occupational performance. Examples of activities that occupational therapy may address are shopping at the local market, putting on a coat for going outdoors, using the computer to write an email to the grandchildren, and organizing the household. In early stages of PD, patients' occupational therapy goals often include enhancing 'normal' activity performance, and prevention of giving up activities and roles. In later stages of PD this will shift towards enabling adapted involvement in valued activities. The role of occupational therapy extends to enabling primary caregivers to support and supervise the patient in daily activities whilst considering their own well-being.<sup>1, 53, 61</sup>

A theoretical model illustrating the focus of occupational therapy is the Person-Environment-Occupation model (PEO model). This model describes occupational performance as the outcome of the dynamic interdependence of three elements: the person (mind, body and spiritual qualities; performance skills), the environment (physical, social, cultural and spiritual context) and occupation (groups of meaningful tasks and activities).<sup>63, 64</sup>

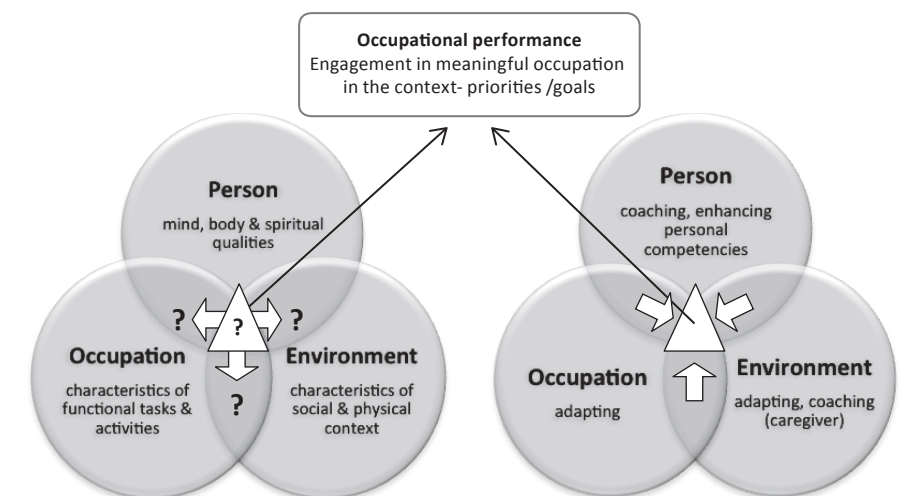
Occupational performance is optimal when there is a maximal fit between the personal resources and competencies, the environmental supports and barriers, and the occupational demands.<sup>63, 64</sup> Occupational therapists are experts in analysing occupational performance and the interacting characteristics of the person, occupation and environment that hamper or support occupational performance (Figure 2). Subsequently, the occupational therapist enable the person to create a better fit between the three elements and thus optimise occupational performance (Figure 3).

Hereby, occupational therapists tailor the strategies to the goals, preferences and resources of the person with PD and the caregivers. In sum, occupational therapists provide a person-centred approach in the domain of occupational performance.

In the field of PD, the unique role of occupational therapists is recognised in multidisciplinary guidelines and models of care, but scientific evidence to support or refute the contribution of occupational therapists is lacking.<sup>59, 62</sup>

### Available evidence and person-centred occupational therapy guidelines

Up to 2008, the content of occupational therapy in the field of PD care had primarily been based on the expertise of individual occupational therapists.<sup>65, 66</sup> Consequently, there was a lack of clarity and a wide variance in occupational therapy interventions. This compromised the quality of care. Therefore, under the auspices of "Ergotherapie Nederland" (the Dutch occupational therapy association), a primary working group was appointed to develop a national clinical guidelines for occupational therapy in PD care.<sup>61</sup> A systematic method of evidence-based guideline development of the Dutch Institute of Healthcare Improvement (CBO) and international standards of guideline evaluation were used.<sup>67</sup> The ICF model and PEO model underpin the theoretical framework of the guidelines. Accordingly, the guidelines support a person-centred and occupation based approach. Due to a scarcity of studies specifically evaluating



**Figure 3** Occupational therapy assessment (left) and Occupational therapy interventions (right)  
Based on the Person-Environment-Occupation (PEO) Model<sup>63, 64</sup>

occupational therapy in PD, the recommendations in the guidelines are based on evidence from related specialties in combination with expert consensus and field testing rounds. It resulted in a document with graded practice recommendations for: (1) referral indications, (2) person-centred and occupation based assessment methods, and (3) various strategies (mainly compensatory) to enable activity performance and participation.<sup>61</sup> The focus is on addressing the needs of people with PD, as well as the needs of their primary caregivers.

The guidelines promote a comprehensive person-centred occupational therapy assessment in order to: (1) understand the person's occupational identity and coping; (2) identify and prioritise the most important problems in occupational performance that the person is eager to reduce; (3) analyse which factors of the person, the occupation or the environment hinder or support the patient's occupational performance. To this end, the guidelines provide specific recommendations for methods of assessing the person with PD and the partner/caregiver.<sup>61</sup> These include methods of narrative occupational history taking, a standardised measure to set and rate priorities, an activity log, standardised observation of occupational performance, and an environmental checklist. Because of the potential influence of environmental factors, assessment preferably takes place in the natural performance context (e.g. at home).

If needed, the occupational therapist seeks relevant information from other disciplines involved to obtain a comprehensive impression of the patient. The occupational therapist concludes the assessment phase with a collaborative goal setting and treatment planning session. The choice of intervention strategies is tailored to fit the goals and preferences of the person, and the opportunities for change in the personal competencies and resources, the environmental demands and supports and the demands of the activity. Often a combination of different strategies is required to maximise the Person-Environment-Occupation fit for occupational performance goals. Details on recommended occupational therapy interventions for PD can be accessed in the guidelines.<sup>61</sup> An example of one goal and subsequent intervention is described in Box 2.

The Dutch guidelines document was published in 2008 and is the basis of the training of ParkinsonNet occupational therapists.<sup>61</sup> In 2011, an English translation of the guidelines became freely available online.<sup>68</sup> Still, there was no evidence for the effectiveness of occupational therapy according to these guidelines. This is the reason for initiating the studies described in this thesis.

## Box 2 Case study

Mrs. H is 67 years old and lives with her partner in a two-room bungalow. She has had Parkinson's disease for 7 years and is in the moderate disease stage.

### Priority

Struggling to cook a meal

### Problem analysis

When cooking a meal Mrs. H experiences incidental freezing in the crowded kitchen when turning to gather items. Due to slowed movement and reduced mental flexibility she has difficulty to manage multitasking and to handle the time pressure induced by the cooking task. As a result not all dishes are ready simultaneously, after cooking the kitchen is a chaos and Mrs. H feels exhausted. She no longer enjoys cooking and her partner suggests it might be better to buy readymade meals. Mrs. H does not want to give up cooking and is eager to find ways of better managing the activity.

### Goal

I enjoy cooking a simple two-person hot meal (maximum 2 pots) four days a week.

### Intervention/strategies

After explaining and discussing the options with Mrs. and Mr. H, the following strategies are agreed upon and employed:

#### Person:

Mrs. H learns to apply a structured planning strategy (cognitive compensatory strategy) for preparing meals to reduce time pressure and multitasking. She performs some preparation tasks earlier in the day. She learns to use a high stool at the kitchen sink to sit down when preparing vegetables. This prevents the need for dual motor tasking (i.e. maintaining balance while rinsing or cutting) and allows her to focus on the fine motor task. The physiotherapist noted that Mrs. H responds well to the 'marching cue' and the auditory cue. In the kitchen Mrs. H prefers to use the marching cue and the occupational therapists trains the use of this cue during a cooking task.

Social environment: Now both partners have insight into the contributing problems to the difficulty in cooking, the partner is advised to support his wife in her performance by allowing her to take sufficient time and by avoiding introducing extra tasks while she is cooking (e.g. no conversation).

Environmental adaptation: the small kitchen table is put with one end to the wall to create more space. Items in the cupboards are rearranged to reduce the number of required turns. A suitable stool is placed at easy access for the kitchen sink.

Occupation: The cooking task is simplified by performing the separate steps/tasks in a sequence (reduced multitasking) and by using some ready peeled potatoes and cut vegetable mixes. The frequency of preparing a fresh meal is reduced from 7 to 4 times. By cooking larger portions the meals can be divided over the 7 days.

## Outcome measurement in occupational therapy

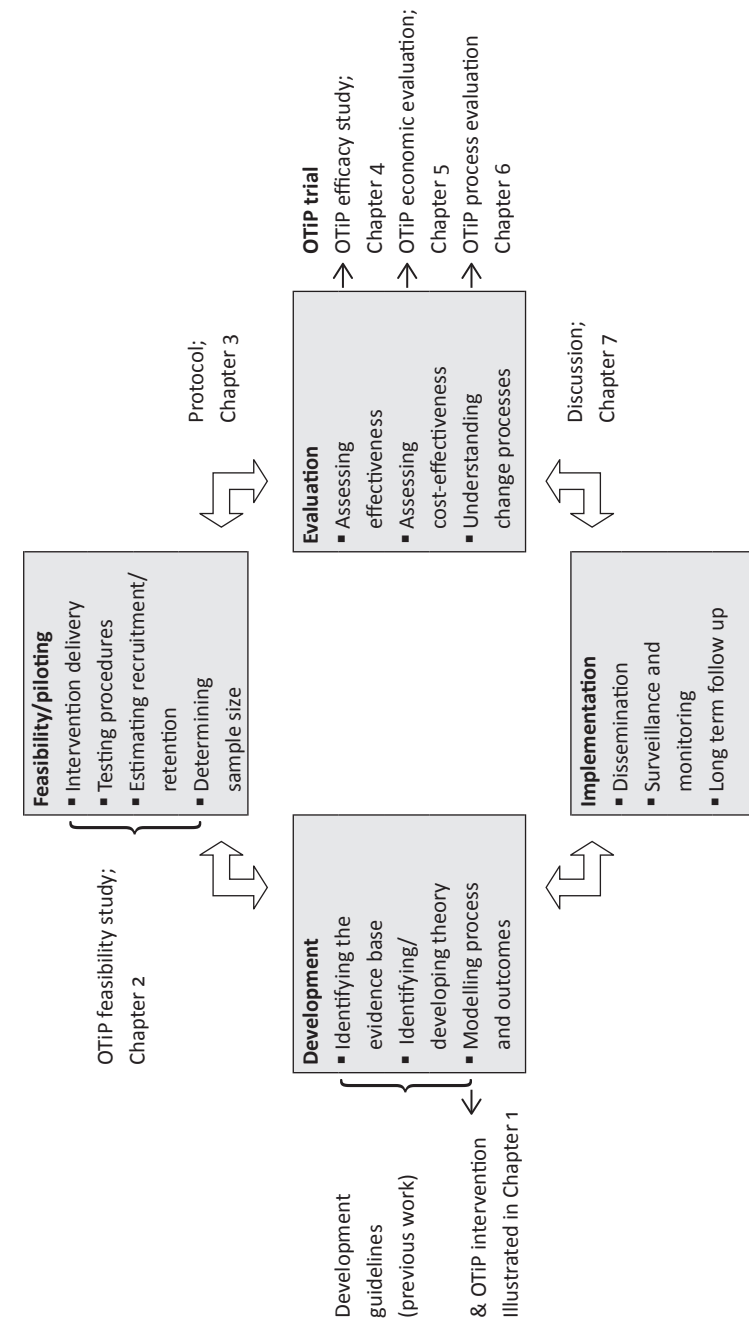
Occupational therapy as described above contains multiple interacting intervention components, varying goals and tailored strategies. Therefore, it is a highly complex intervention.<sup>5, 69</sup> One of the challenges in evaluating complex and individualised interventions is selecting an appropriate outcome measure to determine efficacy.<sup>70, 71</sup> The domain of the outcome needs to be meaningful to the patient. Additionally, it needs to closely reflect the targeted goals of the intervention and be able to capture change. A complicating issue is that individualised goals and tailored interventions lead to varied outcomes. Especially when the domain of the goals and the expected outcome are connected to occupational performance like in occupational therapy. For example, for person A the goal might be to be able to look after the grandchildren; for person B to be able to visit a museum. Hence, we expected that standardised questionnaires evaluating a fixed list of activities would not be comprehensive nor specific enough to measure the effect of occupational therapy. The evaluation and selection of appropriate outcome measures was therefore an important part of the feasibility/piloting phase of our study.

## Aims and Outline of this thesis

The aim of this thesis was to assess the (cost)effectiveness of home-based occupational therapy on daily functioning of persons with PD and their caregivers. We also endeavored to gain insight into the actual treatment delivery and recipients' and therapists' experiences with the intervention. It is obvious that occupational therapy needs more research. With the results of this thesis we want to contribute to the evidence base underlying occupational therapy and provide suggestions for further development and implementation of occupational therapy interventions for people with PD.

The outline of this thesis follows the phased design of our research project which was inspired by the Medical Research Council's framework for the development and evaluation of complex interventions (Figure 4). This framework propagates a systematic and phased approach with attention for modelling and exploratory testing before conducting a definite evaluation and implementation.<sup>4, 5</sup>

The intervention we planned to assess was a home-based occupational therapy intervention according to the existing guidelines. Therefore the *development phase* of this intervention had mainly taken place in the previous project of developing the guidelines. To arrive at the study OTiP-intervention protocol we standardised the intervention process, added tools to summarise the assessment phase and to select strategies, and we provided a structured case notes format.



**Figure 4** Occupational therapy in Parkinson's disease (OTiP) study in relation to key elements of the development and evaluation process according to the Medical Research Council framework

The *feasibility/piloting phase* is described in **Chapter 2**. This presents the results of the exploratory randomised controlled trial with a mixed methods design. We assessed the feasibility of study procedures (recruitment/retention, burden, adherence to the protocol), the intervention delivery (intervention fidelity), the experiences with the intervention (patients/caregivers) and the potential impact of the intervention. The results were used to fine tune the OTiP intervention and the study procedures and to decide upon the most appropriate outcome measures.

Based on the outcomes of chapter 2, **Chapter 3** describes the design of the subsequent definite randomised controlled trial, including the evaluation of efficacy, cost effectiveness and process in detail.

**Chapter 4** reports the results of this randomised controlled trial in the *evaluation phase*: the OTiP efficacy study. The trial profile and primary and secondary outcomes are presented and discussed.

In **Chapter 5** the results of the OTiP study's economic evaluation from a societal perspective are presented. It addresses cost differences between the intervention and control group and the intervention's net monetary benefit.

**Chapter 6** provides a detailed analysis of the process of the occupational therapy intervention as delivered in the trial. In this study we aimed to get insight into (1) the treatment fidelity: the dose, the protocol process adherence, and content of treatment delivered compared to expected based on the protocol; (2) the level treatment enactment by recipients (patients and caregivers); (3) The experiences of recipients with the intervention process and outcomes; (4) The experiences of therapists on perceived benefit of the intervention for the recipients and on the barriers and facilitators for successful treatment delivery.

**Chapter 7** is the general discussion of this thesis. We reflect on the overall findings of the studies and provide suggestions for clinical practice and for future research in the field of occupational therapy in PD care.

Finally, **Chapter 8** (English) and **Chapter 9** (Dutch) summarise this thesis.

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## Chapter 2

### **The impact of occupational therapy in Parkinson's disease: a randomised controlled feasibility study**



**Published as**

Sturkenboom IH, Graff MJ, Borm GF, Veenhuizen Y, Bloem BR, Munneke M, Nijhuis-van der Sanden MW. The impact of occupational therapy in Parkinson's disease: a randomized controlled feasibility study. *Clinical Rehabilitation*. 2013;27(2):99-112.

## Abstract

**Aim:** To evaluate the feasibility of a randomised controlled trial including process and potential impact of occupational therapy in Parkinson's disease (PD).

**Design:** Process and outcome were quantitatively and qualitatively evaluated in an exploratory multicentre, two-armed randomised controlled trial at three months.

**Participants:** Forty-three community-dwelling PD patients and difficulties in daily activities, their primary caregivers and seven occupational therapists.

**Intervention:** Ten weeks of home-based occupational therapy according to the Dutch guidelines of occupational therapy in PD versus no occupational therapy in the control group.

**Main measures:** Process evaluation measured accrual, drop-out, intervention delivery and protocol adherence. Primary outcome measures of patients assessed daily functioning: Canadian Occupational Performance Measure (COPM) and Assessment of Motor and Process Skills. Primary outcome for caregivers was caregiver burden: Zarit Burden Inventory. Participants' perspectives of the intervention were explored using questionnaires and in-depth interviews.

**Results:** Inclusion was 23% (43/189), drop-out 7% (3/43) and unblinding of assessors 33% (13/40). Full intervention protocol adherence was 74% (20/27), but only 60% (71/119) of baseline COPM priorities were addressed in the intervention. The outcome measures revealed negligible to small effects in favour of the intervention group. Almost all patients and caregivers of the intervention group were satisfied with the results. They perceived: 'more grip on the situation' and used 'practical advices that make life easier'. Therapists were satisfied, but wished for a longer intervention period.

**Conclusions:** The positive perceived impact of occupational therapy warrants a large-scale trial. Adaptations in instructions and training are needed to use the COPM as primary outcome measure.

## Introduction

Parkinson's disease (PD) is a neurodegenerative disease affecting both motor and non-motor brain systems, which can result in multiple deficits like impairments in gait, balance, hand coordination, memory or executive functioning. This inevitably leads to various limitations in daily functioning and increasing need for support.<sup>1-4</sup> Consequently, PD has a great impact on quality of life of patients and their caregivers.<sup>5-7</sup> A client-centred and multidisciplinary approach in Parkinson's care is required to address the great variety of difficulties and needs of patients.<sup>8-9</sup>

Occupational therapy aims to optimise a person's functional performance and engagement in everyday activities and life roles and uses a client-centred approach.<sup>10</sup> Although the merits of occupational therapy for persons with PD are widely recognised by clinicians and a few small scale studies appear promising, rigorous studies testing the specific effects of client-centred occupational therapy in persons with PD do not exist.<sup>11-14</sup>

An important obstacle in setting up clinical trials was a lack of best practice guidelines for occupational therapy in PD.<sup>15-16</sup> In 2008, we developed national Dutch guidelines for occupational therapy in PD to improve uniformity and quality of care. The guidelines are based on (1) extensive literature review in the fields of occupational therapy and rehabilitation for people with PD and other neurodegenerative conditions, (2) expert opinion and (3) field-testing rounds.<sup>8,17</sup> This resulted in 31 recommendations for referral, assessment and treatment of PD patients and their caregivers. Implementation is facilitated by using the guidelines as a basis in the training of occupational therapists who join the Dutch multidisciplinary networks of professionals specialised in PD (ParkinsonNet).<sup>18-19</sup> We do not know, however, what the effectiveness is of occupational therapy according to these guidelines within the context of multidisciplinary care.

Following the recommendations of the Medical Research Council for evaluation of complex interventions, we conducted a phase II exploratory study using a combination of quantitative and qualitative methods, as a precursor for a definite trial.<sup>20-21</sup> The aim of this study, called the OTiP pilot, was to evaluate the feasibility of a randomised controlled trial including process of the study procedures (accrual, drop-out, burden, protocol adherence), process of the intervention (actual intervention delivery, OTiP intervention adherence and experiences) and the potential impact of occupational therapy in PD.

## Methods

An exploratory randomised controlled trial was performed with random allocation of intervention 2:1 control, and an assessor-blinded post-intervention measurement at three months followed by a qualitative evaluation of the intervention procedures and outcome. The qualitative evaluation had a phenomenological design exploring perceptions of the intervention procedures and outcome using individual interactive interviews with participants (patients, caregivers, therapists), and focus group discussion with therapists.

Full ethical approval was granted by the medical ethical committee of Arnhem-Nijmegen and the trial was registered at [clinicaltrials.gov](https://clinicaltrials.gov) (identifier NCT01010529).

From October 2009 to February 2010 we recruited patients from four neurology outpatient departments in different regions in the Netherlands. Eligible patients were diagnosed with idiopathic PD, lived at home, reported difficulties in daily activities relevant for the patient (covering self-care, domestic activities, work or leisure), and had a non-professional caregiver who *could* provide assistance for at least twice a week. Exclusion criteria were: use of occupational therapy in the last 12 months, disabling comorbidity, inability to complete questionnaires (i.e. due to severe cognitive problems), and participation in another intervention trial. Considering the complexity of the intervention and potentially large heterogeneity in participants, we expected to require approximately 40 patient-caregiver dyads in the exploratory randomised controlled trial to gain insight into all relevant feasibility factors. This is not based on a power calculation.

The neurologist sent invitation letters to PD patients who had visited the outpatient department in the previous year. The researcher (IS) or research assistant (both assessors) informed interested patients and their caregivers by phone and verified eligibility. Accordingly, the assessors visited eligible patient-caregiver dyads to obtain written consent and perform baseline measurements. After baseline, an independent secretary executed a balanced randomization per region using a computerized randomization protocol.

To provide the intervention we recruited one to two occupational therapists in each participating region. They had to be part of ParkinsonNet, because these therapists already completed three days training in Parkinson's disease and the occupational therapy guidelines. To further improve intervention skills and to inform about trial procedures, study participants received an additional three days training. In the qualitative evaluation we included all participating therapists and their first willing patient-caregiver dyad.

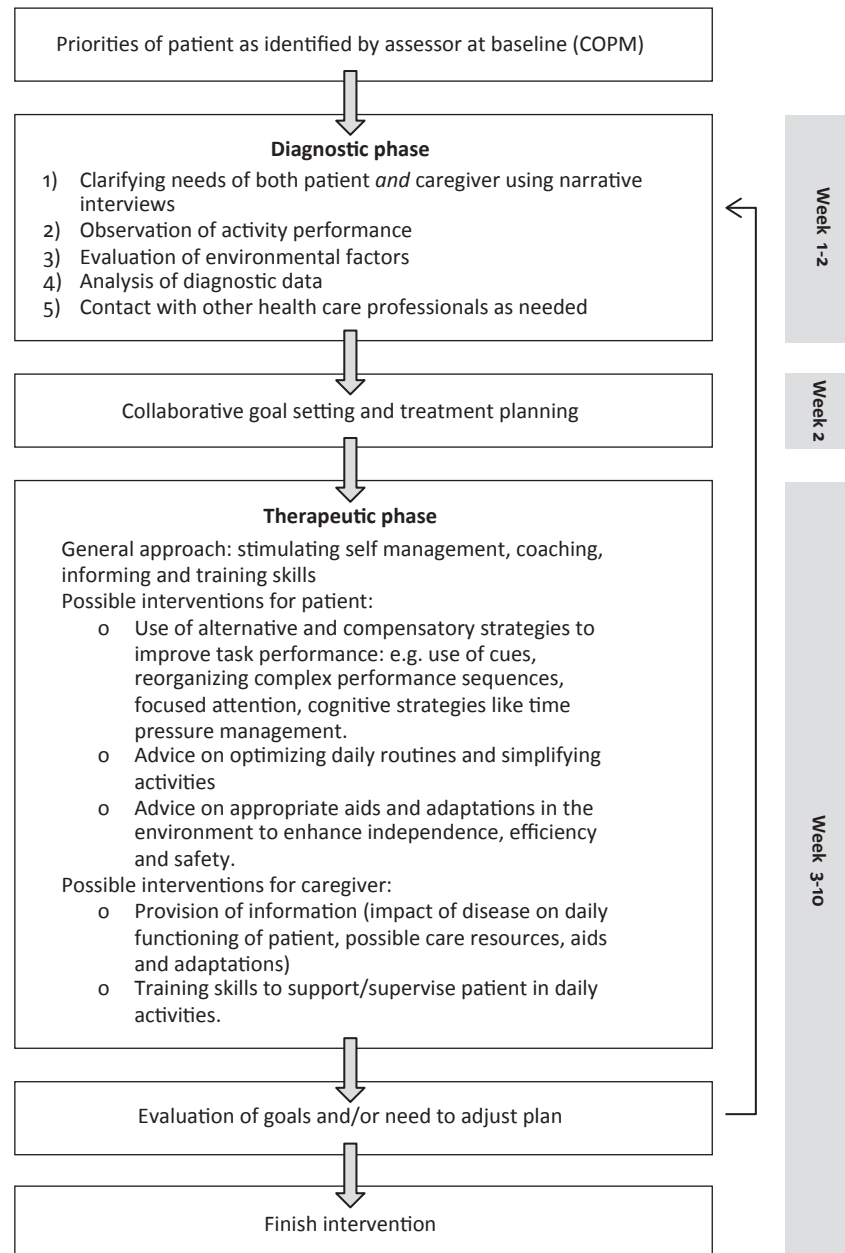
The intervention group received occupational therapy following the Dutch guidelines of occupational therapy in PD.<sup>17</sup> The intervention process was standardized and described diagnostics, goal setting and a guide for choosing appropriate individualised interventions for patients and caregivers. Priorities in problems in daily functioning were evaluated at baseline by the assessor using the Canadian Occupational Performance Measure (COPM),<sup>22</sup> and the named activities and scores were communicated to the therapist by an independent secretary. The therapist formulated the treatment plan based on this information and additional information from the therapist's own diagnostic phase. The intervention was delivered at home for 10-weeks within three months while the number of sessions could vary depending on complexity of goals, with a maximum of 16 sessions (45-60 minutes). The characteristics of the OTiP intervention are summarised in Figure 1. The control group did not receive any occupational therapy intervention until after the three months measurement (wait-listed).

In both groups, patients and caregivers could have other medical or allied healthcare interventions except occupational therapy during the study period. Patients and caregivers registered these other interventions in a care utilisation questionnaire at baseline and three months, both focusing on the preceding three months (not reported).

A variety of assessment procedures and measures were used. To enable evaluation of baseline characteristics, assessors collected data on the patients' and caregivers' age, gender, caregiver's relationship to the patient, severity of Parkinson's disease motor symptoms (Unified Parkinson Disease Rating Scale-part III<sup>23</sup> and Hoehn &Yahr), duration of PD, comorbidity (Cumulative Illness Rating Scale-Geriatrics<sup>24-25</sup>) and baseline scores on the outcome measures. From the therapists we collected years of work experience and years of involvement in ParkinsonNet.

Trial recruitment was evaluated based on inclusion and dropout rates. Assessment procedures were evaluated based on patients' and caregivers' feedback on clarity and perceived burden of the assessments using custom made questionnaires at baseline and post-intervention. Assessors used an assessment log to register duration of the visit, adherence to the assessment protocol and any irregularities encountered. To evaluate the feasibility of the intervention we used a number of measurements. First, therapists completed standardised OTiP patient records allowing insight into adherence to the OTiP intervention and actual treatment delivery (process, content and time). We compared priorities identified by patients in the baseline COPM, with goals addressed in the intervention. Second, within one month after the intervention we evaluated perceived barriers and facilitators for a successful intervention by individually interviewing the participants of the qualitative evaluation. The interviews were conducted by a trained research assistant (YV) not involved in the randomised controlled trial. Topics were: expectations beforehand, experiences of the intervention procedures and outcome, personal context and factors of the therapist. In addition,





**Figure 1** Characteristics of the OTiP intervention. COPM, Canadian Occupational Performance Measure

all patients and caregivers of the intervention group received a custom made questionnaire based on the Consumer Quality index<sup>26-27</sup> evaluating the experiences of the intervention (OTiP-experiences questionnaire). Third, after completion of all interventions the researcher (IS) and interviewer (YV) evaluated the therapists' opinions on adherence to and practicability of the OTiP-intervention protocol and the standardised patient records in a focus group discussion.

For outcome measurement, assessors conducted assessments at baseline (before randomization) and at three months (post-intervention) at the patients' homes. To allow for possible response fluctuations, live performance measures were administered within 1-2 hours after medication intake (the *on* phase).

Primary outcome for patients was daily functioning, encompassing both objective evaluation of skills with the Assessment of Motor and Process Skills (AMPS)<sup>28</sup> and patient-rated perceived performance in activities measured with the COPM.<sup>22</sup> These are both well established generic occupational therapy measures with good clinometric properties.<sup>28,29-33</sup> Both measures produce two scores and in our study protocol we selected one score from each measure as primary outcome. In the COPM we used the perceived performance score as primary outcome.

Although the second score of satisfaction with performance is important, we primarily wanted to know whether occupational therapy contributes to improved performance in daily activities as perceived by the patient. In AMPS we used the score of process skills as primary outcome and not motor skills. This was because process skills incorporate skills of adaptation to problems encountered in the performance of activities and this is the specific focus of occupational therapy in the multidisciplinary context. The primary outcome for caregivers - perceived burden - was measured using the Zarit Burden Inventory (ZBI).<sup>34</sup> For detailed characteristics of the primary outcome measures see Table 1.

The impact of the intervention was also evaluated in the OTiP-experiences questionnaire<sup>26-27</sup> with specific questions on perceived benefit of the intervention, and in the in-depth interviews.

Descriptive statistics were used to present baseline characteristics and results of quantitative process measurements. Primary outcome measures were analysed by covariance analysis with post-intervention scores at three months as dependent variable and baseline scores as covariates. Two-sided 95% confidence intervals were calculated, as well as effect size (Cohen's *d*).

For the analysis of qualitative evaluation data, we used the constant comparative method resulting in identification of main themes.<sup>35</sup> Triangulation was performed with data from interviews, questionnaires, and patient records.

**Table 1** Primary outcome measures: properties

Measure	Domain	Type	Outcome range
<b>Patient</b>			
Canadian Occupational Performance Measure (COPM) performance	Perceived performance capacity in 3–5 self-prioritized problems in daily activities	Semi-structured interview	Mean score range: 1–10 (higher scores indicate better skills). Clinical important difference 2
Assessment of Motor and Process Scale (AMPS) process skills	Quality of process performance skills in daily activities	Rater observation	Linear score range: –3 to 4 (higher scores indicate better skills). For the process skills there is a cut-off point of 1, below which assistance in daily activities is to be expected. Clinical important difference 0.5
<b>Caregiver</b>			
Zarit Burden Inventory (ZBI)	Perceived burden of care. Covering competence, feelings of anger, embarrassment and the impact of care giving on social relations, other responsibilities, health, privacy, time for self and finances	Self-report questionnaire	Item 0–4 Sum score range: 0–88 0–20 indicates no to mild burden; 21–40 mild to moderate burden and above 40 high burden. Clinical important difference: not available

## Results

Forty three patient–caregiver dyads were included (n= 29 patient–caregiver dyads in the intervention group; n=14 in the control group) in the randomised controlled trial (Figure 2). Of the 43 patient-caregiver dyads, three dyads and one extra caregiver dropped out because of worsening co morbidity or hospitalisation of patient or caregiver. Seven female occupational therapists with a mean experience of 16.6 years (range 6–26 years) participated in the trial. In the qualitative evaluation all seven therapists and their first willing patient-caregiver dyads participated (21 individuals). The intervention group and control group were well matched at baseline (Table 2) and did not differ in primary outcome scores (Table 4; first columns). Analysis of type

**Table 2** Baseline characteristics

	Intervention n =27	Control n =13
Age (years): mean (SD)	66.7 (11.8)	68.5 (9.6)
Men/women: n	19/8	10/3
Living status with caregiver/alone: n	26/1	11/2
Paid work: n	4	1
Time since diagnosis (years): mean (SD)	7.5 (7.1)	6.9 (4.3)
Hoehn & Yahr stage: n		
1	4	0
2	15	11
3	7	2
4	0	0
5 <sup>a</sup>	1	0
UPDRS III, motor function, range 0–108 <sup>a</sup> : mean (SD)	28.2 (12.7)	28.5 (10.3)
CIRS-G, comorbidity range 0–56 <sup>a</sup> : mean (SD)	9.3 (2.8)	10.2 (4.4)
MMSE, cognition, range 0–30 <sup>b</sup> : mean (SD)	28.2 (1.9)	27.3 (1.4)
Use of physiotherapy, yes/no: n	16/11	8/5
Age caregiver (years): mean (SD)	61.5 (16.8)	62.5 (6.4)
Caregiver, men/women: n	7/19	3/10

<sup>a</sup>Minimum score indicates best functioning. <sup>b</sup>Maximum score indicates best functioning. UPDRS III, Unified Parkinson Disease Rating Scale part III; CIRS-G, Cumulative Illness Rating Scale-Geriatrics; MMSE, Mini-Mental State Examination.

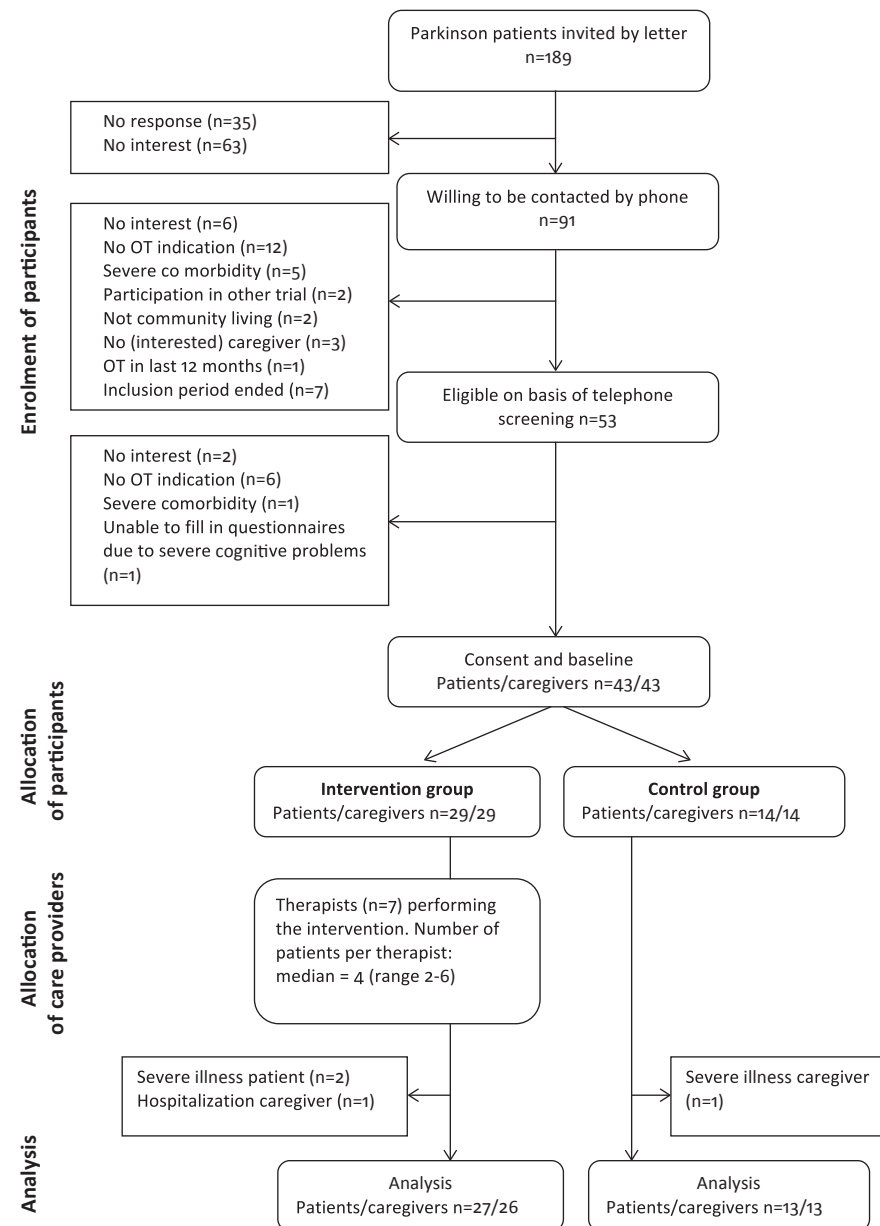
and frequency of priorities in daily functioning identified in the baseline COPM (total 178) reveals that patients' priorities covered domains of self-care/housekeeping (55%; 98), leisure/day structure (40%; 71) and work (5%; 9).

Priorities involved improving activity performance and performance skills (ease, speed, safety) (77%; 137), taking up (new) activities (8%; 14), dealing with fatigue (8%; 14), and improving feeling of self-efficacy in daily functioning (7%; 13).

### Process evaluation

To include 43 dyads, 189 patients were approached, giving an inclusion percentage of 23%. Drop-out was 7% (n=3). Participants notified no adverse events, clear procedures and no burden. Although we instructed participants not to reveal group allocation to the assessor, blinding was broken unintentionally in 13 of 40 analyzed dyads (33%).

Analysis of the OTiP-patient records show that therapists performed all sub processes of the OTiP intervention in 74% (n=20) of patient–caregiver dyads. Adherence was



**Figure 2** Recruitment and Participant Flow Randomised Controlled Trial.  
OT, occupational therapy

lowest in the sub processes of formulating treatment goals *together* with the patient and caregiver (adherence 81%; n=22) and conducting a separate caregiver interview (adherence 81%; n = 22). The therapists addressed 60% (71/114) of patients' priorities formulated in the baseline COPM. In six patients (22%) *all* baseline COPM priorities were addressed and in 13 patients (48%) problems not identified in the baseline COPM were (also) addressed.

None of the therapists used the maximum intervention dose: average number of sessions was 7.9 (SD = 2.40), mean patients' face-to-face contact was 6.6 hours (SD = 2.07) and 2.4 hours for caregivers (SD = 2.16). For five out of 27 patient-caregiver dyads the occupational therapy intervention was not terminated at 10 weeks.

Results from the interviews on perceived facilitators and barriers of the intervention are presented in Table 3. Although expectations at the beginning differed, patients and caregivers valued various aspects of the intervention positively, namely: the thorough diagnostic phase, the client-centred approach, involvement of the caregiver, and the home setting. Expertise of the therapist and personal click were deemed important, as well as unity and sharing between patient and caregiver. Therapists mentioned a lack of willingness or ability to change behaviour (skills or routines) in some patients as a barrier. The duration of 10 weeks was mentioned by therapists and some patients and caregivers as too short to achieve optimal results.

In the OTiP experiences questionnaires all patients and caregivers of the intervention group indicated the expertise of their occupational therapist as adequate and stated that they had been involved in joint goal setting. Most patients (76%; 19/25) and caregivers (71%; 17/24) judged 10 weeks intervention as adequate, although five patients and caregivers mentioned this had been too short, and the remaining one patient and two caregivers found it too long. Two patients and caregivers in the intervention group had not completed this question.

In the focus group discussion, therapists expressed appreciation for the structure of the OTiP intervention, although they found the diagnostic phase too lengthy for some patients. Initially they feared they were not being client-centred when not immediately starting therapy. They indicated that it took a while to get used to taking the time for a structured diagnostic phase in which client-centred problems and goals could be explored. Therefore they stressed the importance of adequate training and practice. Again, all therapists mentioned that the period of 10 weeks was too short for interventions on behavioural changes and for delivery of particular aids and adaptations.

### Outcome evaluation

Analysis of the primary outcome measures at three months demonstrated a small positive trend in favour of the intervention group. Results are presented in Table 4. As to be expected in an underpowered pilot study, differences were not significant

**Table 3** Qualitative Evaluation of the Intervention (n=21)

Theme	Subthemes	Example quotes
<b>Results</b>		
'More grip on the situation'	Improved understanding: 'What balance is all about' 'That this is a consequence of Parkinson's' 'That he can actually do it himself' 'Possibilities in the healthcare resources'	'...the insight. Of what makes up the strain and how I can better influence that, how I can better balance it. That helped me a lot.' (patient) 'Mentally that had a very positive effect. Because you [herself] get grip of the situation and the feeling of yes he [patient] can do it, although just in another way.'(caregiver) 'That he accepts that there are days that performance is more difficult'(therapist)
'Practical advices that make life easier'	'Carrying things out in a different way'	'... occupational therapy has contributed to adaptations that make life more pleasant and easier.'(patient) 'For me it has had the result that now he [patient] does those little things himself.'(caregiver) 'She can help him out of bed in a less straining way and negotiate with him what he can and can't do himself.' (therapist)
<b>Possible barriers and facilitators</b>		
Expectations	'A question mark' and 'openness towards the intervention'	'...but with a question mark. Am I in a phase that that [OT] can contribute?'(patient) 'So let it go, we'll see what comes. That in itself I found quite pleasant'(caregiver) '...I don't know whether I have something to offer to this man' (therapist)
Diagnostic phase	'Long and difficult & setting out a good line'	'It gives a basis of trust and contact. You are being heard. So because of that I think I become opener, because she listens.' (patient) 'I was quickly tempted to address the practical issues and had to sit on my hands: no, just continue on this part and let the conversation flow.'(therapist)
Duration	'Just enough' versus 'too short'	'The duration was appropriate, so that was good. At one point, yeah, it was no longer needed that she came.'(patient) 'For us it was too short. Considering the situation it could have been longer. The results for us are only in the long term.'(caregiver) '[if it was not for the intervention protocol] Then I would have given him more time and now it was my time I had to force upon him.' (therapist)
Involvement caregiver	'We're going through it together'	'Also for her [caregiver] process, I think. She has to start realise as well what it [PD] all involves. We both don't know this.'(patient) 'That [involvement in intervention] I found not more than normal. You are husband and wife. And especially these sort of things you have to do together.' (caregiver) 'And when I told him [patient] what was important for her [caregiver], in a neat way. Now then he was more conscious of oh it is not only for me important but also for her.' (therapist)
Home-setting	'My home is where I do the things'	'In the hospital you are in a theoretical situation, while my problems are here [home]. So then she can better see what it looks like here and how we can adapt things than there [hospital].'(patient) 'I found that [treatment at home] real good...I believe that there you can pick up certain things best.' (caregiver) 'Yes I find that [treatment at home] very good. Because people will tell you more how things are and they can show how they can do other things.' (therapist)

**Table 3** Continued

Theme	Subthemes	Example quotes
Factors of the therapist	'There has to be a click' 'Address what I want' 'The expertise of the therapist'	'Attention for me as a person, looking at the situation, adjusting interventions to the situation, practical, thinking creatively to find what fits best in that situation.' (patient) Yes, there was a click, so then it becomes a lot easier.' (caregiver) I think I was able in the right way to make him think [about issues]. That is the feeling he gave me." (therapist)
Factors of the patient/ context	Openness for changes  Possibility to change Level of sharing between patient and caregiver.	'In the process of acceptance you can ask yourself a hundred thousand times why me or why not and how. ...I gain more from looking at what are we going to do.' (patient) 'And we are like 'if they want to help you, you have to go for it.' (caregiver) 'I think the fact that a lot has happened in his life and was still going on, I think that has had a lot of influence. So I think that the results are affected by that, he cannot do everything at once.' (therapist)

**Table 4** Results primary outcome measures

Measure	Baseline intervention (SD)	Baseline control (SD)	Three months intervention (SD)	Three months control (SD)	Mean difference intervention versus control corrected for baseline (95% CI)	Effect size (d)
COPM, performance	4.31 (1.50)	4.64 (0.92)	5.36 (1.57)	5.20 (1.01)	0.36 (-0.48-1.91)	0.23
AMPS, process skills	0.82 (0.44)	1.00 (0.59)	0.99 (0.55)	1.08 (0.46)	0.05 (-0.18-0.28)	0.11
ZBI	15.95 (11.30)	17.69 (14.64)	17.25 (11.32)	19.84 (15.24)	-0.84 (-4.31-2.64)	0.07

COPM, Canadian Occupational Performance Measure; AMPS, Assessment of Motor and Process Skills; ZBI, Zarit Burden Inventory; SD, standard deviation; CI, confidence interval; *d*, Cohen's *d* for effect size.

( $P > 0.05$ ) and within-group variability was high. Following Cohen's rule for interpreting effect sizes, only the COPM reached a small effect size.<sup>36</sup> Almost all patients (96%; 23/24; three missing responses) and caregivers (96%; 24/25; one missing response) indicated in the OTIP experiences questionnaire that they were mostly or absolutely satisfied with the results of the intervention. The mean grade for satisfaction with the intervention was 8.5 on a (scale of 1–10). Also in the interviews, participants reported benefits. Even patients with mild problems were pleasantly surprised about what the occupational therapy intervention had offered them. The perceived results were diverse, but two main themes were identified: 'more grip on the situation' and 'practical advices that make life easier' (Table 3).

## Discussion

This study confirms the value of a phase II exploratory study as recommended by the Medical Research Council. What we learned is that although recruitment and assessment procedures were feasible, only 23% of patients met the inclusion criteria of the study. This means that we need a large PD population to recruit an appropriate number of patients (estimated  $n = 168$ ;  $\alpha = .05$ , power 80%) for the final randomised controlled trial. Therapists mentioned they needed time and coaching to get confidence and competence in conducting the intervention according to protocol. And although only a small effect size on the COPM and negligibly small effect sizes on the AMPS and ZBI were found, the interviews and OTIP experiences questionnaires clearly revealed a positive impact of the occupational therapy intervention. We will next discuss some of the findings in detail and their implications for modifications of a future trial.

A relevant finding was that not all priorities in daily functioning identified at baseline in the COPM, were addressed in the intervention. Reason for this could be that during the intervention patients opinions on priorities might change. This was also found in test-retest studies of the COPM.<sup>30, 32</sup> Therapists could base treatment planning on both the COPM conducted by the assessor and findings from the therapist's own diagnostic phase which also included evaluation of priorities. Evidence suggests that the therapist's personal attitude, skills and expertise can also greatly influence the priorities and choice of goals or the focus in the intervention.<sup>32, 37-38</sup> To optimise a client-centred and focussed treatment planning process, more attention should be paid in the training and coaching of therapists to the translation of COPM priorities to Specific, Measurable, Attainable, Relevant and Time-based goals and treatment plans *together* with the patient. Moreover, in the administration protocol of the baseline COPM an extra verification of priorities can be included. Following a client-centred approach, we do not expect nor demand that all priorities of the baseline COPM are addressed in each patient, but with suggested adaptations congruence between baseline COPM priorities, goals and interventions is likely to improve. During coaching divergence from the baseline priorities can be monitored and should be justified by the therapist.

Although participants appreciated the intervention process and content, opinions varied on the duration. Interestingly, especially the therapists perceived 10 weeks as too short to fully address some goals and felt a time pressure. In contrast, they did not use all hours allowed. Therapists felt they could not increase the intensity as patients needed time for information to settle and for trying out advices. Nonetheless, there is evidence in PD rehabilitation that high intensity training during a short period is more effective for skill acquisition.<sup>8, 14</sup> It seems logical that this might also be the case for occupational therapy interventions that involve skill training. However, most occupational therapists working in the community, are not used to provide intensive interventions in a short period of time, and it seems that the issue of intensity needs to be addressed in the training of therapists. Enhancing and consolidating behaviour change in the longer term requires an early focus on patient-caregiver dyad empowerment using own resources (self management). Only in incidental cases of lengthy application procedures for aids and adaptations, a follow up visit after delivery will be necessary to ensure safe and proper use of the equipment.

Although we did not intend to provide definite data on effectiveness, we expected larger changes in the outcome measures. A likely explanation for the small effect size on the COPM is the already mentioned incongruence between baseline COPM and focus of the intervention. Despite this drawback, we still judge this client-centred measure to be suitable as a primary outcome measure for evaluating the occupational therapy intervention following the Dutch guidelines. Standardised measures of limitations in activities do not cover the diversity and specificity of types

of priorities worked on in occupational therapy.<sup>31</sup> In other studies the COPM was successfully used as a primary outcome measure for client-centred occupational therapy interventions.<sup>39-40</sup> We expect that the earlier suggested adaptations in the administration protocol and coaching for therapists will increase the responsiveness of the COPM.

Lack of responsiveness of the AMPS might be explained by the high mean process skills score at baseline suggesting a ceiling effect for participants who scored little inefficiency. Moreover, for some patients problems with daily functioning were exclusive to the *off* phase and the AMPS observation took place in the *on* phase. Finally, the AMPS is an observational performance measure which only evaluates motor and process skills and does not capture effects on interventions addressing feeling of self-efficacy, taking up activities or overall problems such as fatigue. In a definite trial, we would therefore suggest the use of an observational measure of activity performance and additional measures of participation and fatigue as secondary outcomes.

For caregivers, the small effect size in the ZBI might be explained by low mean caregiver burden at baseline. Caregivers' own needs were therefore less important than we had expected beforehand. As participants valued the caregivers' involvement in the occupational therapy intervention positively, we recommend keeping the intervention protocol the same, but evaluating caregiver burden as secondary outcome with subgroup analysis for caregivers' perceived burden.

Another feasibility issue the study revealed was the high unblinding rate. It is difficult to prevent unblinding as patients are visited at home and asked to talk about their daily lives. In a future trial unblinding rates might be improved by instructing patients to only give answers to the questions and not elaborate on the context of their answers. A researcher who is not the assessor should be readily available by phone to answer any questions from participants.

A weakness of the study was that all therapists treated a relatively small number of patients each. A large amount of therapists was chosen in order to be able to cover a large geographical region and to make it easier to generalise the results to clinical practice. However, this limits a rapid increase in expertise and gives rise to more variation in treatment delivery and outcome. In a future large scale study we still suggest using a high number of therapists, but from the current findings we recommend providing regular coaching and monitoring of therapists *during* the study period to improve adherence to the OTiP intervention. The allowance of variation in amount and content of treatment sessions can be seen as a limitation in the study protocol but is a consequence of following a client-centred approach. As providing adequate intensity seemed to be an issue in this exploratory trial, the dose might need to be a factor to control for in a definite trial.

In summary, this feasibility study pointed out that it is warranted to test the effectiveness of occupational therapy following the Dutch guidelines in a future trial,

but outcome measures need to be refined, treatment intensity should be increased, and aligning priorities with goals and interventions together with the patients and caregivers is an important issue in training and coaching of therapists. Moreover, a planned process evaluation alongside the effect study is appropriate to enable conclusions to be drawn.

### Clinical messages

- Patients with Parkinson's disease and caregivers report positive benefits from the home-based occupational therapy intervention, but effectiveness is not yet ascertained.
- During the intervention process, incongruence can arise between initial priorities and intervention goals. This may affect the responsiveness of a client-centred outcome measure like the Canadian Occupational Performance Measure.

### Acknowledgements

We like to thank Anita de Vries-Uiterweerd, research assistant, for her contribution in the data collection; Jolanda Gons (Verpleeghuis Birkhoven), Irma Jansen (Ziekenhuis Rijnstate), Rianne Merks and Selma Sari (Ergotherapie Thuis), Astrid Rothuis (Ergotherapie Dieren), Martine Valk (Ziekenhuis Rivierenland) and Silvia van Rijswijk (Stichting Zorgcentra de Betuwe) for delivering the OTiP- intervention; the local coordinators of the participating hospitals: Tanja Hoogendoorn (Ziekenhuis Rivierenland), Edo Hoogerwaard (Ziekenhuis Rijnstate), Ad Hovestadt (Meander Medisch Centrum), Erik van Wensen (Gelre Ziekenhuizen), for the collaboration in this study; and all participating patients with Parkinson's disease and their caregivers. Clinical Trial Registration: NCT01010529.

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## Chapter 3

### **Effectiveness of occupational therapy in Parkinson's disease: study protocol for a randomised controlled trial**



**Published as**

Sturkenboom IH, Graff MJ, Borm GF, Adang EM, Nijhuis-van der Sanden MW, Bloem BR, Munneke M. Effectiveness of occupational therapy in Parkinson's disease: study protocol for a randomized controlled trial. *Trials*. 2013;14:34.

## Abstract

**Background:** Occupational therapists may have an added value in the care of patients with Parkinson's disease (PD) whose daily functioning is compromised, as well as for their immediate caregivers. Evidence for this added value is inconclusive due to a lack of rigorous studies. The aim of this trial is to evaluate the (cost) effectiveness of occupational therapy in improving daily functioning of PD patients.

**Methods/Design:** A multicenter, assessor-blinded, two-armed randomised controlled clinical trial will be conducted, with evaluations at three and six months. One hundred ninety-two home-dwelling PD patients and with an occupational therapy indication will be assigned to the experimental group or to the control group (2:1). Patients and their caregivers in the experimental group will receive ten weeks of home-based occupational therapy according to recent Dutch guidelines. The intervention will be delivered by occupational therapists who have been specifically trained to treat patients according to these guidelines. Participants in the control group will not receive occupational therapy during the study period. The primary outcome for the patient is self-perceived daily functioning at three months, assessed with the Canadian Occupational Performance Measure. Secondary patient-related outcomes include: objective performance of daily activities, self-perceived satisfaction with performance in daily activities, participation, impact of fatigue, proactive coping skills, health-related quality of life, overall quality of life, health-related costs, and effectiveness at six months. All outcomes at the caregiver level will be secondary and will include self-perceived burden of care, objective burden of care, proactive coping skills, overall quality of life, and care-related costs. Effectiveness will be evaluated using a covariance analysis of the difference in outcome at three months. An economic evaluation from a societal perspective will be conducted, as well as a process evaluation.

**Discussion:** This is the first large-scale trial specifically evaluating occupational therapy in PD. It is expected to generate important new information about the possible added value of occupational therapy on daily functioning of PD patients.

## Background

Parkinson's disease (PD) is the second most common neurodegenerative disorder. It is a complex disease affecting both motor and non-motor systems in the brain. As a result patients can have a wide range of deficits in performance components, including mobility, balance, hand dexterity, memory and executive functioning. As the disease progresses, effectiveness of the medication regime often decreases, and daily functioning and social participation become increasingly compromised.<sup>1-3</sup> PD has a great impact on the quality of life of both patients and their informal caregivers.<sup>4-6</sup> The costs of care are high, partly due to the increasing need of support.<sup>4</sup> Improvement of quality of life and reduction of healthcare costs might be achieved by maintaining or improving the patient's skills and independence in daily activities, and also by reducing caregiver's burden. To address the great variety of needs in a complex and progressive disease like PD, a client-centred and multidisciplinary approach is required.<sup>7-9</sup>

Within multidisciplinary care for PD patients, the primary role of occupational therapy is to optimise activity performance and engagement in valued activities and roles in the home or community context (occupational performance). The contribution of occupational therapy in Parkinson's is widely recognised, but systematic reviews reveal a lack of rigorous studies to draw conclusions on the effectiveness of occupational therapy in PD care.<sup>10-12</sup> Some studies evaluate occupational therapy as part of a multidisciplinary intervention,<sup>13-17</sup> but the specific contribution and added value of occupational therapy cannot be determined from these studies.

From 2006 to 2008 we developed guidelines for occupational therapy in PD (in Dutch), under the auspices of the Dutch Association of Occupational Therapy with the aim to improve uniformity and quality of occupational therapy in PD.<sup>18,19</sup> The guidelines cover specific methods for occupation-based assessment of patients and their caregivers and self-management and compensatory strategies to maintain or enhance occupational performance or occupational performance patterns in daily life.

Our hypothesis is that occupational therapy according to the Dutch guidelines has an added value within multidisciplinary care for PD patients and their caregivers. We expect that addressing the complex occupational performance issues from an occupational therapy perspective will improve daily functioning, more so than if occupational therapy is not involved. Improved daily functioning will result in enhanced participation in daily activities among patients, reduced caregiver burden, an improved quality of life for both patients and caregivers, and a reduction in costs for society. To test this hypothesis, we followed the steps of the framework for evaluation of complex interventions of the Medical Research Council.<sup>20,21</sup> Based on a phase II exploratory trial,<sup>22</sup> we have improved the procedures for the currently proposed randomised controlled trial (phase III trial). This trial, the OTiP study,

evaluates the effectiveness and cost-effectiveness of occupational therapy according to the Dutch guidelines for occupational therapy in PD.

## Methods/Design

### Trial design

A multicenter, assessor-blinded, two-armed randomised controlled clinical trial will be conducted. Patients and their caregivers will be assigned to the experimental group or to the control group in a ratio of 2:1, respectively. This way the patients have twice as much chance to be in the intervention than in the control group. This ratio will enhance the inclusion, whereas there will hardly be any power loss compared with a 1:1 randomization. Randomization will be based on a computerized minimization algorithm with the following minimization factors: baseline primary outcome measure (Canadian Occupational Performance Measure (COPM) performance:  $<5$ ;  $\geq 5$ ), severity of disease (Hoehn and Yahr (H&Y) score:  $<3$ ;  $\geq 3$ ), gender and age group ( $<65$  years;  $\geq 65$  years) of the patient, and patient receiving physiotherapy at baseline (yes/no).

Data on observational and self-reported outcome measures will be collected at baseline, after three months (post-intervention) and after six months (follow-up) (Figure 1). Full ethical approval has been granted by the medical ethical committee of Arnhem-Nijmegen (NL27905.091.09/ABR27905) and the OTiP trial is registered at clinicaltrials.gov (NCT01336127).

### Setting

For inclusion and intervention the study is embedded within ParkinsonNet regions in the Netherlands. ParkinsonNet comprises 65 regional networks of professionals specialised in the treatment of PD patients, and includes a group of specifically trained occupational therapists.<sup>23,24</sup> Ten regional hospitals and 18 occupational therapists in nine selected ParkinsonNet regions agreed to participate. The trial assessments and occupational therapy interventions take place at the patient's home.

### Participants

Eligible patients have idiopathic PD, live at home, and report difficulties in valued daily activities covering the occupational therapy domains of self-care, domestic activities, work or leisure. Exclusion criteria are: occupational therapy intervention in the last three months, predominating disabling co morbidity, and inability to complete questionnaires (that is, due to language problems or a Mini Mental State Examination score  $<24$ ). A primary informal caregiver of each patient can participate in the study when willing and available. Enrolment will take place over a period of 18 months (2011/2012) and procedures are given in Figure 1. Informed consent of patient and caregiver is obtained before the first assessment.

### Intervention

The OTiP intervention protocol follows the principles and recommendations for diagnostics and interventions as described in the Dutch guidelines for occupational therapy in PD.<sup>18,19</sup> The approach is client-centred including shared decision making and supporting self-management of the patient and caregiver in dealing with problems in daily activities. The trial therapist receives the patient's priorities in problems in daily functioning as evaluated at baseline with the COPM.<sup>25</sup>

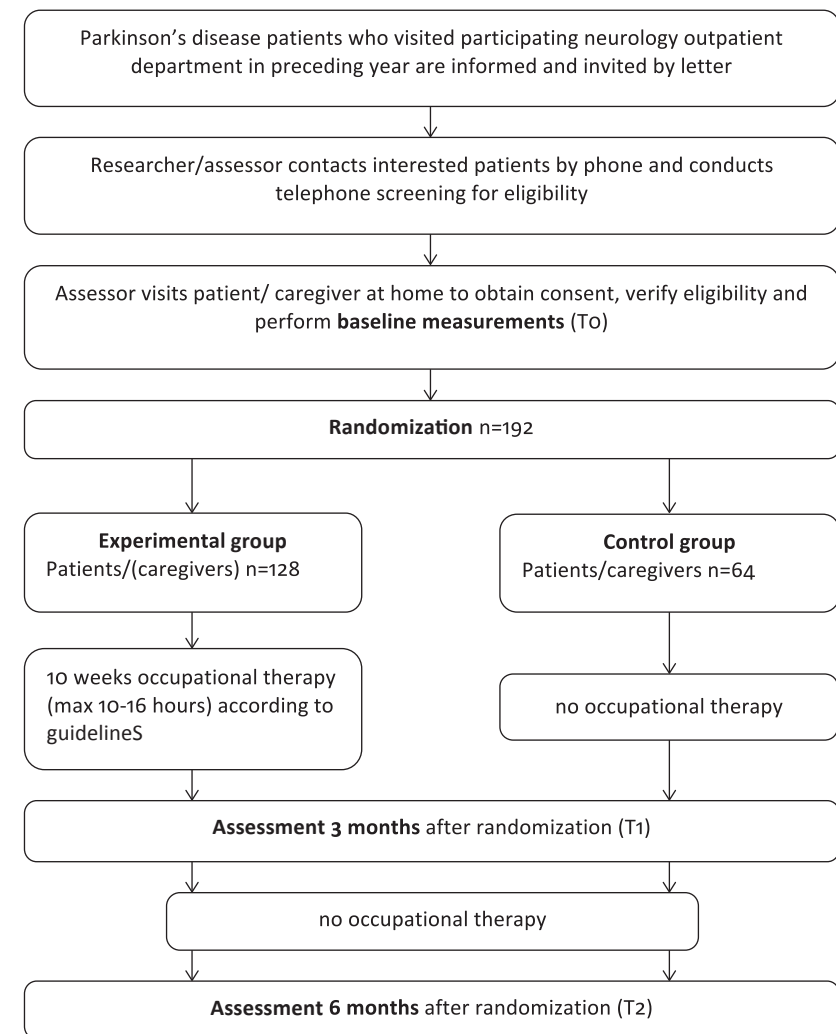
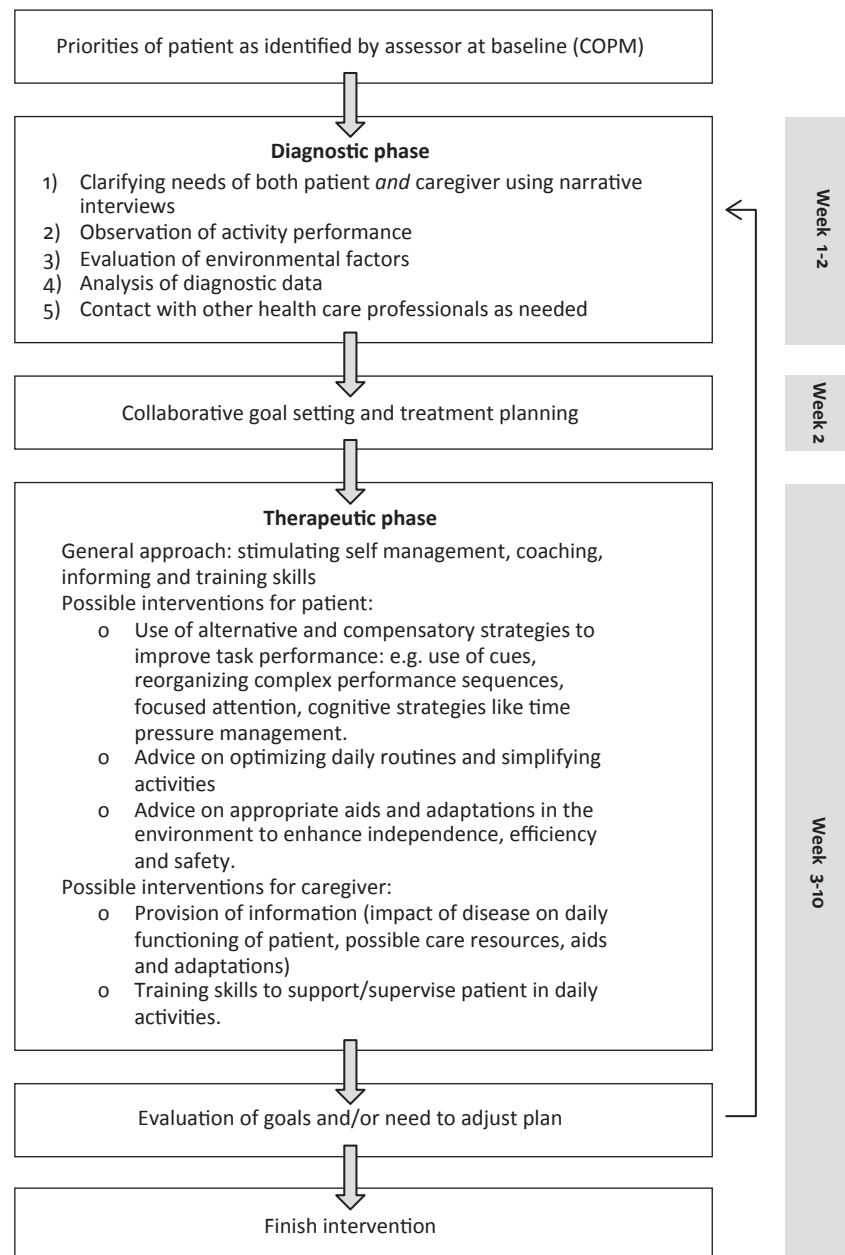


Figure 1 Flow chart of design and enrolment procedures



**Figure 2** Characteristics and process of the intervention

The baseline COPM priorities and additional information from the diagnostic phase shape the treatment plan. The intervention is delivered at the patient's home for a period of ten weeks within three months. Depending on complexity of goals, the amount of sessions can vary with a maximum of ten sessions (only patient goals) or 16 sessions (patient and caregiver goals) of 45 to 60 minutes. Between the three and six month assessments, no occupational therapy will be received. An exception is when in incidental cases of lengthy procedures to apply for aids and adaptations, a follow-up contact after delivery is necessary to ensure safe and proper use of the equipment. Figure 2 summarises the process and characteristics of the OTiP intervention.

The control group does not receive any occupational therapy intervention during the study period (six months). Thereafter, control group patients can receive the OTiP intervention if they wish.

In both groups, patients and caregivers are allowed to receive other medical or allied health care interventions except occupational therapy during the study period. We register the input of other health and social care professionals using a care-utilisation questionnaire at each of the three assessments focusing on the preceding three months.

### Training of trial therapists

Before the start of the trial, the participating occupational therapists follow a three-day training to inform them about the study procedures and to train them to treat the patients and caregivers according to the OTiP-intervention protocol. Special attention is given to enhancing the therapists' skills in coaching and motivational interviewing and in eliciting and collaboratively defining meaningful, individualised goals with the patient and caregiver. Ways to achieve sufficient treatment intensity in ten weeks are discussed. Halfway through the inclusion period a booster training session (one day) is planned. Therapists can use a secure online platform to share issues and experiences and can consult an expert occupational therapist (expertise in the OTiP intervention) to discuss the intervention.

### Assessment procedures

Data from patients and caregivers will be collected at baseline (T0), three months (T1) and six months (T2) by three research assistants (see Table 1). Another eight selected and trained occupational therapists will score the Perceive Recall Plan Perform system (PRPP<sup>26</sup>) in an activity that is video recorded by the assessor. All assessors and PRPP-scorers are blinded for group allocation and each participant will be followed up by the same assessor. Patients and caregivers also fill in self-report questionnaires. Observational tests or measures that follow a semi-structured interview format are conducted in the patient's home environment by the assessor. Considering possible response fluctuations in Parkinson's, measures are administered within one to two

hours after medication intake (the on phase). For budgetary reasons, the six month assessment is conducted by phone and therefore, does not include observational measures.

## Outcome measures

### Primary outcome measure

The primary outcome for effectiveness of this intervention is the patient's self-perceived performance in daily functioning as assessed with the Canadian Occupational Performance Measure (COPM) (see Table 1).<sup>25</sup> The COPM is an individualised outcome measure with a semi-structured interview format and a structured scoring method. The patient identifies three to five activities in which he encounters problems and would like to improve. These activities are subsequently rated by the patient on a scale from 1 to 10 for perceived performance capacity and level of satisfaction with this. The COPM score for performance or satisfaction derives from the mean score of the prioritised activities. Change is evaluated by asking the patient to rescore performance and satisfaction on the original priorities. Studies evaluating the psychometric properties of the COPM (in populations of stroke and various chronic conditions) support the validity and reliability of the COPM.<sup>41-43</sup> Responsiveness for change over three months was established in a population with various conditions, and the results support both criterion and construct responsiveness.<sup>44</sup> There is a high correlation between performance and satisfaction scores.

We selected the COPM as a primary outcome measure in our trial as it fits with the client-centred nature and specific focus of the OTiP intervention. It addresses the patient's priorities and evaluation of valued activities. In the assessment procedures, we specified the COPM administration protocol to improve uniformity in the semi-structured interview and in the formulation of priorities while taking care to maintain the client-centred nature of the instrument. Only the mean performance capacity score will be used as primary outcome.

### Secondary outcome measures

In Table 1 all secondary outcome measures are listed. Patients' secondary outcomes include evaluation of observed performance of daily activities, self-perceived satisfaction with performance in daily activities, participation, impact of fatigue, proactive coping skills, mood, health related quality of life, and overall quality of life. All outcomes at the caregiver level will be secondary and include self-perceived burden of care, objective burden of care, proactive coping skills, mood and overall quality of life. In both patients and caregivers resource use, productivity losses and other costs related to receiving support or providing care are assessed with a questionnaire for the economic evaluation.

## Background variables

Socio-demographic data consisting of age, gender, marital status, education, employment status, and relationship between client and caregiver will be collected at baseline, using a questionnaire. Patient's disease severity will be measured with the Unified Parkinson Disease Rating Scale-part III<sup>45</sup> and with the Hoehn and Yahr (H&Y) scale. The Mini Mental State Examination<sup>46</sup> is used for cognitive screening. Comorbidity is checked with an open question for screening purposes.

## Process measures

To enable explanation of results, process data will be collected. Therapists complete standardised OTiP patient records and a process evaluation summary sheet to provide insight into adherence to the steps of the OTiP intervention and actual treatment delivery (content, amount of sessions and time spent). For each patient seen, the participating trial therapists also record their views of effectiveness of the intervention for the individual patient and caregiver on the process evaluation summary sheet. We will compare priorities identified by patients in the baseline COPM, with goals addressed in the intervention. At the end of the study a focus group will be conducted with all therapists exploring their experiences and views on conducting the OTiP-intervention protocol in daily practice.

Patients and caregivers experiences with the intervention will be evaluated with a custom made questionnaire (OTiP experiences questionnaire) based on the Consumer Quality index.<sup>47,48</sup> It includes mainly closed questions on experiences with the interaction with the therapist, the process and content of the intervention and the perceived effectiveness of the intervention. Assessors register any irregularities in adherence to assessment procedures, including unblinding. Adverse events or irregularities affecting protocol adherence will be registered by the researcher.

## Sample size calculation

In the main study, we intend to enrol 192 PD patients and their caregivers. This is based on the results of the OTiP pilot study with 43 participants. The pilot resulted in a difference of 0.5 in scores on the primary outcome measure (COPM), whereas the standard deviation was 1.35. Based on these assumptions, a t-test would require a control group of 75 patients and an intervention group of 150 patients for 80% power (two-sided testing at 5%). The correlation between baseline and outcome was approximately 0.5 and as a result, the co-variance analysis that is planned only requires a control group of 56 and an intervention group of 112 patients (total of 168).<sup>49</sup> In the feasibility study the dropout rate was 7%. We expect however, that dropout rates will be higher in this main study as the study period is doubled. Therefore we adjust for a 10 to 15% drop-out rate and will include approximately 192 patients.

**Table 1** Outcome measures

Participant	Outcome measure	Instrument	baseline	3 months	6 months
Patient	Self-perceived performance in daily activities <sup>a</sup>	Canadian Occupational Performance Measure (COPM; performance score) <sup>25</sup>	√	√	√
	Self-perceived satisfaction with performance in daily activities	Canadian Occupational Performance Measure (COPM; satisfaction score) <sup>25</sup>	√	√	√
	Objective performance in daily activities	Perceive Recall Plan Perform system (PRPP) <sup>26</sup>	√	√	-
	Participation	Activity Card Sort (ACS) <sup>27,28</sup>	√	√	-
		Utrecht Scale for Evaluation of Rehabilitation Participation (USER-P; satisfaction part) <sup>29,30</sup>	√	√	√
	Health-related quality of life	Parkinson's Disease Questionnaire (PDQ-39) <sup>31-33</sup>	√	√	√
	Impact of fatigue	Fatigue Severity Scale (FSS) <sup>34,35</sup>	√	√	√
	Mood	Becks Depression Inventory (BDI) <sup>36</sup>	√	√	√
	Caregiver	Perceived caregiver burden	Zarit Burden Interview (ZBI) <sup>37</sup>	√	√
Objective caregiver burden		Objective care burden questionnaire; hours of care	√	√	√
Mood		Hospital Anxiety and Depression Scale (HADS) <sup>38</sup>	√	√	√
Both	Quality of life	Euroqol EQ-5D <sup>39</sup>	√	-	√
	Quality of life overall	Visual Analogue Scale for Quality of life; VAS QoL	√	√	√
	Proactive coping	Utrecht Proactive Coping Competence scale <sup>40</sup> (UPCC) <sup>40</sup>	√	√	√
	Resource utilisation	Resource utilisation questionnaire: patient and caregiver version	√	√	√

<sup>a</sup> Primary outcome measure.

## Statistical analysis

### Descriptive statistics

Means, standard deviations and frequencies will be used to describe outcome, background and baseline variables.

### Analysis effectiveness

The primary variable for effectiveness will be analyzed in a covariance model with the COPM scores after three months (T1) as dependent variable. The baseline COPM scores (T0) and the minimization factors will be covariates. Two-sided 95% confidence intervals will be calculated. The analysis follows the principle of intention to treat. Similarly, a secondary analysis will be done evaluating the secondary outcome variables and outcomes for six months. Regarding the caregiver outcomes, we plan a sub analysis for caregivers with low perceived burden at baseline (Zarit Burden Interview (ZBI) ≤20) and high perceived burden of care (ZBI >20).

### Analysis cost-effectiveness

An economic evaluation will be done from a societal perspective by evaluating the differences in total costs in the control and experimental group at three and six months. Total costs include care consumption and productivity loss of patients and caregivers related to Parkinson's and caregiver's hours of care provision to the patient. The number of occupational therapy sessions and total time spent on occupational therapy, will be translated as direct costs of the intervention. Differences in costs between groups over a six-month timeframe will be estimated using regression analysis taking into account potential co-variants. Secondly, utility will be calculated as quality adjusted life year (QALY) over a timeframe of six months using the trapezium rule. QALYs for patients and caregivers are derived from the EuroQol EQ-5D scores using the EQ-5D health tariffs for the Dutch population.<sup>50</sup> Then, cost and QALY differences are combined in an incremental cost-effectiveness ratio (ICER), and using the bootstrap method, confidence intervals surrounding this ICER will be

estimated. We also measure cost-effectiveness by costs per successful treatment. A successful treatment is a treatment with a clinically relevant positive change in the COPM (+2 points) at six months. Reporting the ICER as cost per successful treatment may provide decision-makers with a relatively intuitive means of assessing cost-effectiveness, because the denominator of the incremental ratio is calculated using a clinically meaningful objective.

#### **Analysis process data**

A descriptive analysis will be performed for the quantitative data on the evaluation forms of participants and assessors and the data of therapists of the given intervention. We will analyse the data from the focus group discussion following the constant comparative method.<sup>51</sup>

## **Discussion**

Current evidence for the effectiveness of occupational therapy in PD is scarce and inconclusive. The OTiP trial is the first large-scale randomised control trial evaluating the effectiveness and cost-effectiveness of client-centred occupational therapy in PD. It is difficult to select one comprehensive outcome measure to reflect the effectiveness of a complex intervention that has a broad and individualised scope. We have chosen an outcome measure (namely the COPM) that potentially fits best with the client-centred nature of the OTiP intervention. Additional (secondary) outcomes can be used to capture the multimodal nature of the intervention. For this purpose, we have included a range of secondary outcome measures covering the wide scope of the OTiP intervention.

The main inclusion criteria are self-perceived problems in valued daily activities and the extent to which patients perceive limitations in daily activities or participation restrictions. These inclusion criteria do not always correspond with disease severity or factors like age. Therefore, we expect great diversity in characteristics of our participants and their contexts. This might also result in diversity in outcomes.

To cover the large geographical spread of participants in this multicenter trial, a relatively high number of trial therapists will be involved to deliver the intervention. This means that the average number of patients seen by each therapist within the trial is low. We have taken several measures to enhance and monitor OTiP-intervention protocol adherence, including an online discussion platform and opportunities for coaching during the study.

Recruitment for trials is often difficult. A strength of this trial is the presence of the national ParkinsonNet infrastructure within the Netherlands.<sup>23,24</sup> This allows easier access to neurologists in the participating regional and university hospitals and

their pool of patients. Another important and novel aspect in this study is that all other interventions are allowed to take place during the study. This way, the added value of guideline-based occupational therapy in a usual multidisciplinary care setting can be evaluated. With the comprehensive process evaluation it will also provide information on factors that are important for further improvement of the content or implementation of the guidelines.

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## Chapter 4

### **Efficacy of occupational therapy for patients with Parkinson's disease: a randomised controlled trial**



**Published as**

Sturkenboom IHWM, Graff MJL, Hendriks JCM, Veenhuizen Y, Munneke M, Bloem BR, Nijhuis-van der Sanden MW, for the OTiP study group. Efficacy of occupational therapy for patients with Parkinson's disease: a randomised controlled trial. *Lancet Neurology*. 2014;13(6):557-66.

## Abstract

**Background:** There is insufficient evidence to support use of occupational therapy interventions for patients with Parkinson's disease (PD). We aimed to assess the efficacy of occupational therapy in improving daily activities of patients with Parkinson's disease.

**Methods:** We did a multicentre, assessor-masked, two-armed randomised controlled clinical trial in ten hospitals in nine Dutch regional networks of specialised healthcare professionals (ParkinsonNet), with assessment at 3 months and 6 months. Persons with PD with self-reported difficulties in daily activities were included, along with their primary caregivers. Patients were randomly assigned (2:1) to the intervention or control group by a computer generated minimisation algorithm. The intervention consisted of 10 weeks of home-based occupational therapy according to national practice guidelines; control individuals received usual care with no occupational therapy. The primary outcome was self-perceived performance in daily activities at 3 months, assessed with the Canadian Occupational Performance Measure (COPM; score: 1–10). Data were analysed using linear mixed models for repeated measures (intention-to-treat principle). Assessors monitored safety by asking patients about any unusual health events during the preceding 3 months. This trial is registered with ClinicalTrials.gov, NCT01336127.

**Findings:** Between April 14, 2011, and Nov 2, 2012, 191 patients were randomly assigned to the intervention group (n=124) or the control group (n=67). 117 (94%) of 124 patients in the intervention group and 63 (94%) of 67 in the control group had a participating caregiver. At baseline, the median score on the COPM was 4.3 (IQR 3.5–5.0) in the intervention group and 4.4 (3.8–5.0) in the control group. At 3 months, these scores were 5.8 (5.0–6.4) and 4.6 (3.8–5.5), respectively. The adjusted mean difference in score between groups at 3 months was in favour of the intervention group (1.2; 95% CI 0.8–1.6;  $p < 0.0001$ ). There were no adverse events associated with the study.

**Interpretation:** Home-based, individualised occupational therapy led to an improvement in self-perceived performance in daily activities in persons with PD. Further work should identify which factors related to the patient, environmental context, or therapist might predict which patients are most likely to benefit from occupational therapy.

## Introduction

The progressive disabling nature of Parkinson's disease (PD) increasingly hampers daily activities and social participation.<sup>1,2</sup> The diversity and complexity of needs of patients with PD and their caregivers warrant a patient-centred and multidisciplinary care approach.<sup>3–5</sup> Within this approach, medical management is complemented with input from allied health professionals who focus on the impact of PD on daily functioning. The evidence is strongest for physical therapy, which focuses on mobility-related functions and activities.<sup>6</sup> By contrast, occupational therapy focuses primarily on supporting participation in daily life, enabling the patient to engage in roles that are meaningful to him or her, and to optimise activities in the domains of self-care, leisure, household and work-related activities.<sup>3,7</sup> The potential role of occupational therapy for management of PD is recognised in multidisciplinary guidelines,<sup>5,8,9</sup> but its use is not supported by evidence owing to an absence of well-designed and properly powered trials.<sup>10,11</sup> Findings from two pilot studies suggested that occupational therapy might support a better functioning of PD patients in daily activities.<sup>12,13</sup> In another pilot study in patients with multiple system atrophy, individualised occupational therapy improved daily activities and quality of life.<sup>14</sup> Some large randomised controlled trials involving PD patients have assessed multidisciplinary interventions that included occupational therapy,<sup>15–17</sup> but the specific contribution of occupational therapy was not assessed.

This scarcity of evidence probably explains the limited use of occupational therapy in the management of PD. Findings from a UK-based surveys in 1995 suggested that 13–25% of patients were referred to occupational therapy.<sup>18</sup> In a Dutch survey in 2004, only 9% of patients consulted an occupational therapist.<sup>19</sup> With the increased attention to multidisciplinary care, these rates might be expected to have risen since, but in a recent Dutch trial in 2013,<sup>20</sup> use of occupational therapy was still only 8% over an 8-month period in areas that offered usual care. Another issue is the timing of referral: an audit of services in the UK<sup>21</sup> showed that the mean time to first referral to occupational therapy is 6 years, suggesting that any potential role of occupational therapy in prevention of functional decline in early PD is not used fully.

In the Netherlands, we addressed these issues by developing practice guidelines for occupational therapy in PD using evidence from related specialties, combined with expert opinion.<sup>22</sup> We used these guidelines in clinical practice to train occupational therapists who take part in multidisciplinary networks of healthcare professionals specialised in PD treatment (ParkinsonNet).<sup>23–25</sup> This process helped to harmonise clinical practice and served to standardise the intervention within clinical trials.

In a phase 2 exploratory trial, we assessed the feasibility of the occupational therapy intervention and explored its clinical effect in the context of the Dutch ParkinsonNet model.<sup>12</sup> The results of this pilot study justified a large trial (the

Occupational Therapy in Parkinson's disease [OTiP] trial), with some adjustments to the protocol from that of the pilot study. We report findings from the OTiP trial, in which we examined the efficacy of occupational therapy according to Dutch practice guidelines. We hypothesised that this intervention would improve perceived performance of PD patients in daily activities compared with usual care. We also expected greater participation in daily activities by patients and lower caregiver burden, leading to improved quality of life for both patients and caregivers.

## Methods

### Participants

We did a multicentre, assessor-masked, randomised controlled clinical trial with 3 and 6 months follow-up within the context of ParkinsonNet.<sup>23-25</sup> The trial protocol, approved by the medical ethical committee of Arnhem-Nijmegen (NL27905.091.09/ABR27905), has been outlined previously.<sup>25</sup>

Patients with a diagnosis of PD according to the UK Brain Bank criteria<sup>27</sup> at ten hospitals (in nine ParkinsonNet regions) were invited by letter to participate. Two occupational therapists (IHWMS and YV) phoned interested patients to provide additional information about the trial and interviewed them for initial screening of eligibility for inclusion. Eligibility criteria were: living at home and reporting difficulties in meaningful daily activities (i.e. activities that patients needed or wanted to do) — an indication for occupational therapy). We excluded patients with a diagnosis of atypical parkinsonism and those who had received occupational therapy in the preceding 3 months, had predominant disabling comorbidity, had insufficient understanding of the Dutch language or had a Mini- Mental State Examination score of less than 24. The patient's primary informal caregiver also participated when willing and available. Patients and caregivers gave written informed consent at enrolment. Sociodemographic data of patients and caregivers and data on disease severity were collected at baseline.

### Randomisation and masking

After baseline assessment, patients were randomly assigned, stratified by region to the intervention or the control group 2:1 by using a computer-generated minimisation algorithm. Factors that were expected to affect the outcome were selected for minimization, namely improvement potential (PD severity, indexed by Hoehn and Yahr score <3 vs ≥3, and baseline perceived performance in daily activities, measured by the Canadian Occupational Performance Measure (COPM) score <5 vs. ≥5), expected variance in nature of daily activities by sex and age (<65 years vs ≥65 years), and receipt of physiotherapy at baseline (yes vs no).

Assessors were masked to treatment allocation. Patients and therapists could not be masked, but participants (i.e. patients and their caregivers) were urged not to discuss their allocation status with their assessor. At the assessments at 3 months and 6 months, the assessors recorded whether their masking was broken.

### Procedures

Within 2 weeks after randomisation, the experimental group received 10 weeks of home-based occupational therapy according to the Dutch guidelines of occupational therapy in PD.<sup>22</sup> Interventions included advice or strategy training in activities, or adaptations of tasks, daily routines or environment (i.e. assistive devices; Figure 1). In the OTiP intervention, the caregivers' needs in supporting the patient in daily activities (e.g. when and how to assist in activities) were also assessed, and addressed if needed. The mix of intervention strategies used was individually tailored to alleviate the problems in activities prioritised by the patient and to suit the patient's coping style, the patient's capacity to change, and the environmental and social context in which the targeted activity is usually done (appendix).

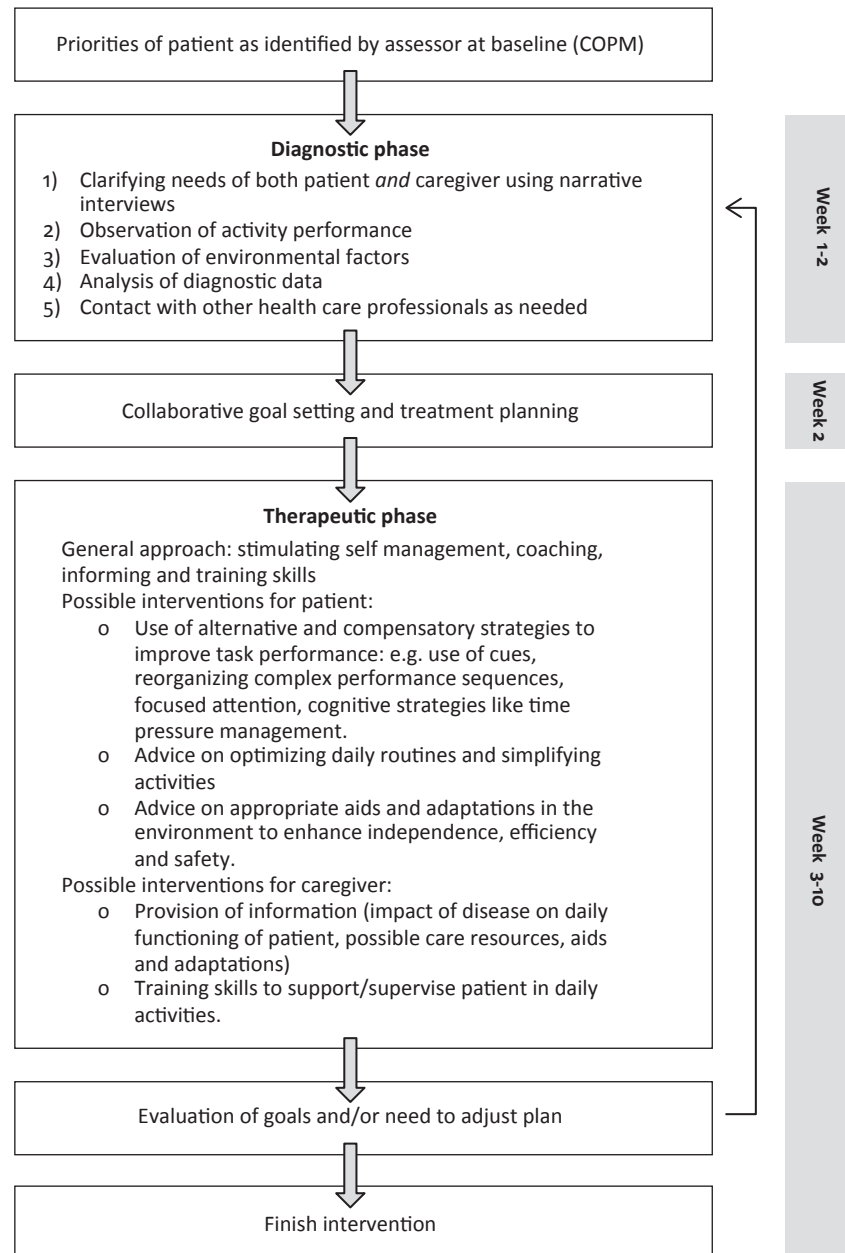
Depending on the complexity of issues to be addressed, the number of sessions could vary, but with a maximum of 16 h over the 10 weeks. Session lengths could also vary, but were mostly 1 h. The control group did not receive occupational therapy during the study. Patients and caregivers in both groups were allowed to receive other medical, psychosocial, or allied healthcare interventions.

Eighteen occupational therapists delivered the intervention. As part of their ParkinsonNet membership, all therapists had received at least 3 days of training in PD treatment according to practice guidelines. The participating therapists were all women, with a median practice experience of 12 years (range 2–28) years and a median ParkinsonNet experience of 2 (range 1–4). OTiP therapists received 3 days of additional training before the start of the study, and a 1-day booster training halfway through the study. To discuss issues and experiences, therapists could use a secure online platform and consult an expert occupational therapist (IHWMS).

Details on treatment delivery were collected by scoring all patient records using predefined process indicators: the content of all delivered treatments was analysed with respect to the extent to which individual steps in the OTiP-intervention protocol had been followed (adherence: 0–100%), and the level to which the intervention addressed the activities prioritised at baseline (COPM congruence: 0–100%).

The medical ethical committee identified no foreseeable risks associated with the intervention. Nonetheless, assessors monitored safety at the time of each assessment, asking about any unusual health events during the preceding 3 months.

Assessments of patients and caregivers took place at baseline and at 3 months and 6 months after randomization, and consisted of assessments done by the assessor and questionnaires completed by participants.



**Figure 1** Elements of the OTiP intervention

COPM=Canadian Occupational Performance Measure.

Reproduced with permission from Sturkenboom and colleagues. (2013)<sup>12, 26</sup>

## Outcomes

The primary endpoint was perceived performance in daily activities at 3 months after randomisation, measured with the performance score of the COPM, an individualised outcome measure of daily activities.<sup>28</sup> Through a semi-structured interview, patients identified and prioritised three to five meaningful daily activities in which he or she perceived performance problems to be most salient. Subsequently, patients rated each activity on a 10-point scale for perceived performance capacity (COPM-P; 1=not able to do at all, 10=able to do extremely well) and similarly for performance satisfaction (COPM-S). During follow-up assessment, patients again rated both perceived performance and satisfaction for all activities that were identified at baseline. Clinimetric properties of the COPM have been established in various populations.<sup>29-32</sup> Secondary endpoints for patients included COPM-P score at 6 months, performance satisfaction (measured with the COPM-S), daily activity performance (measured with the Perceive, Recall, Plan, Perform system phase 1), participation in activities (measured with the Activity Card Sort and the Utrecht Scale for Evaluation of Rehabilitation-Participation Satisfaction Scale), effect of fatigue (measured with the Fatigue Severity Scale), proactive coping skills (measured with the Utrecht Proactive Coping Competence Scale), mood (measured with the Beck Depression Inventory), health-related quality of life (measured with the Parkinson's Disease Questionnaire 39 and EuroQol 5 dimensions), and overall quality of life (measured with the Visual Analogue Scale). All patient outcomes were assessed at 3 months and 6 months, apart from the Perceive, Recall, Plan, Perform system phase 1 and the Activity Card Sort, which were assessed at 3 months only. All caregiver outcomes were secondary and were measured at 3 months and 6 months, including self-perceived caregiver burden (measured with the Zarit Burden Interview), amount of care (measured as care minutes per day), proactive coping skills (measured with Utrecht Proactive Coping Competence Scale), mood (measured with the Hospital Anxiety and Depression Scale anxiety and depression subscales), and quality of life (measured with the Visual Analogue Scale and the EuroQol 5 dimensions). We monitored levodopa equivalent dose (LED) and receipt of physical therapy as potential confounding factors. Cost outcomes will be reported elsewhere.

## Statistical analysis

Power calculations were based on our pilot study in 43 PD patients.<sup>12</sup> To achieve a power of 80% on the COPM-P at 3 months, and after adjusting for an expected dropout rate of 10–15%, we aimed to include 192 patients.<sup>26</sup>

We used linear mixed models for repeated measures to study the differences between groups for each of the outcomes. The dependent variable was the outcome measure. The independent fixed variables were group (control and intervention), baseline score, the minimization factors, and the interaction term between measurement

time points and group. Region was treated as a random variable. We present the baseline-adjusted mean difference between groups at each measurement point with 95% CI. The analyses were done using the intention-to-treat principle. We used the Mann Whitney test to measure differences in amount of care delivered by caregivers.

In a post-hoc analysis, we used the Fisher exact test to calculate the proportion of patients in each group who reached a clinically important change (improvement or worsening) on the COPM-P from baseline. The threshold for this minimal clinically important change was defined as a difference of at least 2 points.<sup>28,29</sup> Statistical analyses were done using SAS 9.2 for Windows and SPSS 20 for Windows. The trial is registered with ClinicalTrials.gov, NCT01336127.

### Role of the funding source

This was an investigator-initiated study. The sponsors of the study had no role in study design, data collection, data analysis, data interpretation, or writing of the report. All authors had full access to all the data in the study and agreed with manuscript submission; final responsibility for the decision to submit for publication was taken by BRB, MJLG and MWN-vdS.

## Results

Between April 14 2011 and Nov 2 2012, 1658 patients were informed about the study, of whom 622 were willing to be screened for eligibility (Figure 2). We included the 191 eligible patients, who were randomly assigned 2:1 to the intervention group (n=124) or the control group (n=67; Figure 2). 117 (94%) of 124 of patients in the intervention group and 63 (94%) of 67 in the control group had a participating caregiver.

Baseline characteristics were similar between groups (Table 1). Three patients (2%) in the intervention group and 6 (9%) in the control group dropped out during the study. Of these nine patients, five had Hoehn and Yahr stage 3; the others had milder disease severity. 14 caregivers were lost to follow-up at 6 months (intervention n=4; control n=10), eight of whom perceived low caregiver burden at baseline (Zarit Burden Interview score  $\leq 20$ ).

There were four hospital admissions due to an accident or fall in the intervention group and two in the control group, but these events were judged (by IHWMS) not to be directly associated with the intervention or study procedures. At 3 months' follow-up, masking of assessors was broken in 11 (6%) of 185 cases and at 6 months in seven more cases (18/182 [10%]), owing to unintentional disclosure in 14 (78%) of these 18 cases.

The occupational therapists treated a median of seven PD patients (range three to 11). The mean number of sessions per patient plus caregiver was 8.6 (SD 2.1), and

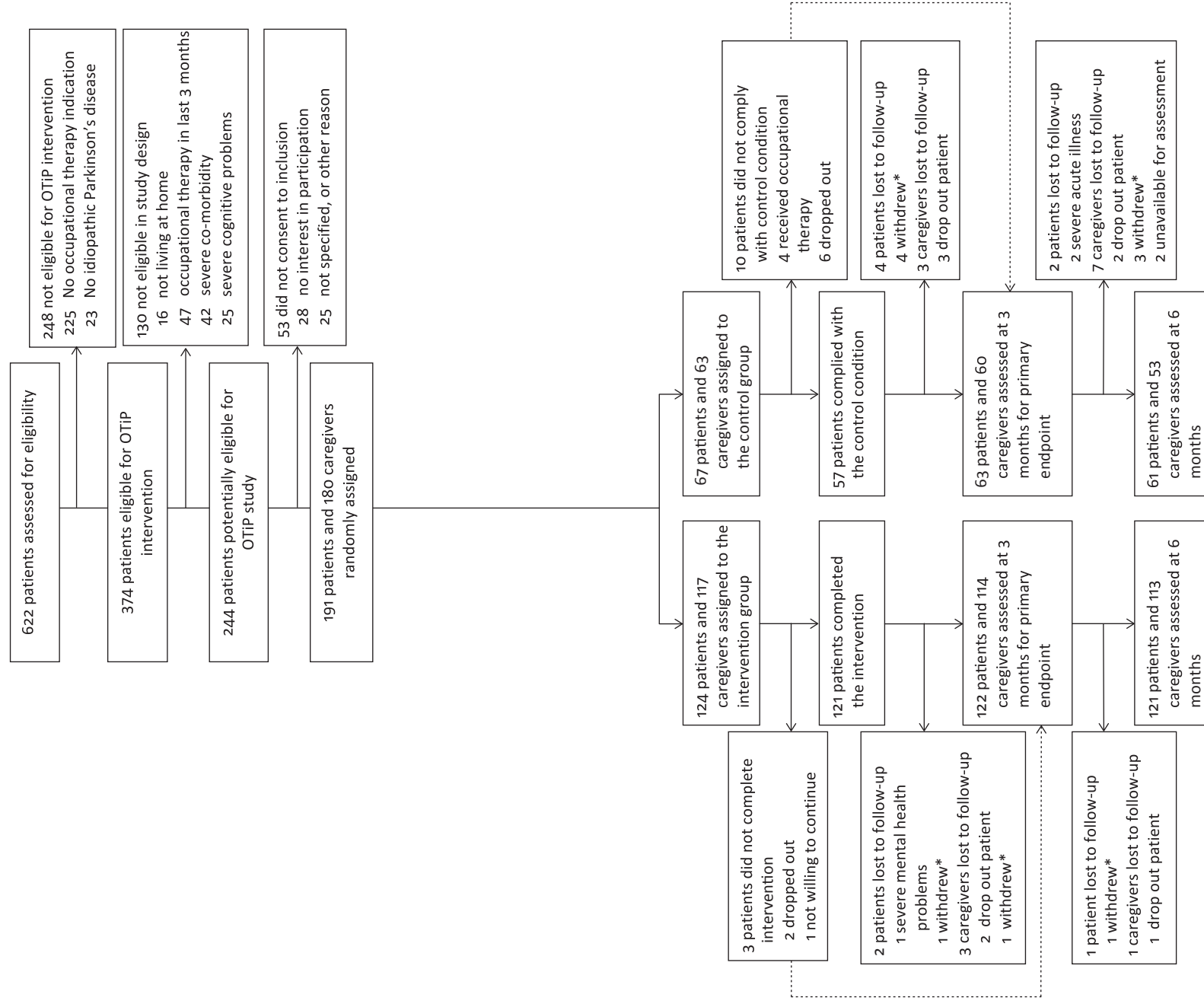
the total mean direct intervention time was 9.4 h (2.3) hours. Mean adherence of therapists to the OTiP-intervention protocol was 94.2% (SD 6.7), and mean congruence between intervention and baseline COPM priorities was 67.4% (20.0). In the intervention group, one patient received additional occupational therapy outside the study after completing the OTiP intervention. 57 (93%) of 61 patients who completed the study, complied with the control condition (no occupational therapy); three patients received occupational therapy after inpatient admission and one via day-care treatment.

At 3 and 6 months, the intervention group had significantly better self-perceived performance on prioritised activities (COPM-P) compared with the control group (both  $p < 0.0001$ ; Table 2). The adjusted mean difference in COPM-P between the intervention group and the control group was 1.2 (95%CI 0.8–1.6) at 3 months (primary endpoint) and 0.9 (0.5–1.3) at 6 months (Figure 3). The COPM differences between groups became significantly smaller over time ( $p = 0.045$ ). Specific PD drug use (LED) was higher in patients in the intervention group than in the control group, but the mean difference between groups was similar at the three points of measurement: baseline 64.1 mg (SE 71.7), 3 months 59.9 mg (81.2), and 6 months 60.9 mg (110.5).

Significant benefits in favour of the intervention were found for satisfaction with performance on prioritised activities (COPM-S; Table 2). There were no other significant differences in secondary endpoints between groups. Patients' satisfaction with the OTiP-intervention at 3 months was good (mean score 8.1 [SD 1.2] on a scale 1–10; data missing for five patients).

Most outcomes for caregivers showed no group differences (Table 3). In a pre-planned sub-analysis assessing the efficacy of the intervention on perceived caregiver burden separately for the groups of caregivers with low burden at baseline (Zarit Burden Interview  $\leq 20$ ) or with high burden (Zarit Burden Interview  $> 20$ ), the mean difference between the intervention and control groups was numerically larger for the caregivers with low burden at both 3 and 6 months. However, group differences at both points of measurement were not significant for either low-burden or high-burden groups. The only caregiver outcome that showed a significant but small effect in favour of the intervention was quality of life at 3 months, measured with the EuroQol 5 dimensions scale ( $p = 0.006$ ). Caregivers' mean grade of satisfaction with the OTiP intervention at 3 months was 7.9 (SD 1.5; data missing for 17 caregivers).

Findings from a post-hoc analysis showed that the proportion of patients attaining a clinically relevant improvement on the COPM-P (increase of  $\geq 2$  points) at 3 months was greater for the intervention group (39/122 [32%]) than for the control group (6/63 [10%]; Fisher's exact  $p = 0.001$ ). The proportion of patients attaining a clinically relevant deterioration (COPM decrease of  $\geq 2$  points) was small in both groups (intervention 1/124 [1%]; controls 2/67 [3%]). Patient demographic characteristics, disease stage, and receipt of physiotherapy were similar between responders (n=39) and non-responders (n=83) in the intervention group (data not shown).



**Figure 2** Trial profile

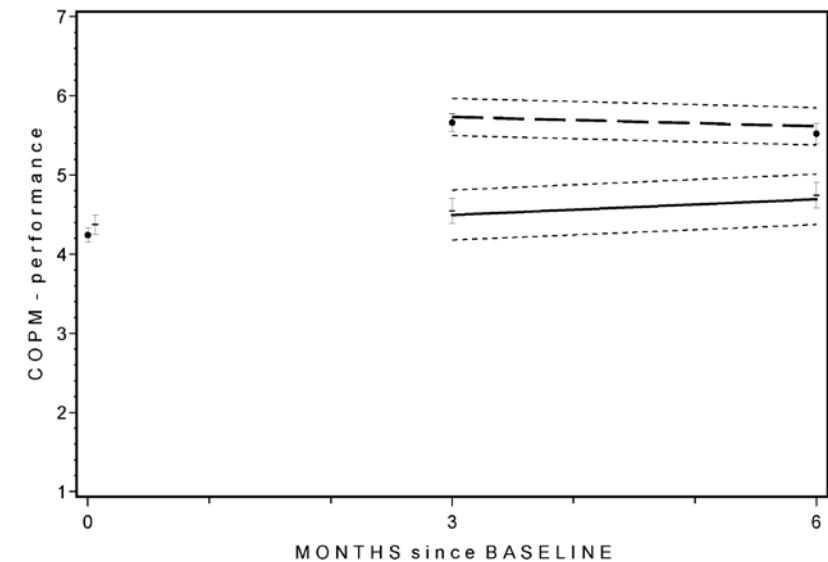
\*No medical reason, but the participant was no longer willing to participate

**Table 1** Demographics and baseline characteristics

	Intervention (n=124 patients, 117 caregivers)	Control (n=67 patients, 63 caregivers)
<b>Patients</b>		
Age (years)	71.0 (63.3–76.0)	70.0 (63.0–75.0)
Sex		
Men	78/124 (63%)	41/67 (61%)
Women	46/124 (37%)	26/67 (39%)
Educational level*		
High	49 /122 (40%)	24/66 (36%)
Middle	45 /122 (37%)	26/66 (39%)
Low	28/122 (23%)	16/66 (24%)
In paid employment	16/124 (13%)	12/66 (18%)
Disease duration (years) †	6.0 (4.0–10.0)	6.0 (3.0–11.0)
Hoehn and Yahr stage		
1	31/124 (25%)	15/67 (22%)
2	46/124 (37%)	32/67 (48%)
3	44/124 (36%)	16/67 (24%)
4	2/124 (2%)	4/67 (6%)
5	1/124 (1%)	0/67 (0%)
UPDRS III (sum) ‡	27 (18.0–36.0)	28 (19.0–36.0)
MMSE (sum)	28 (27.0–29.0)	29 (27.0–29.0)
Daily LED (mg) †	687.5 (415.5–957.7)	550.0 (332.5–1033.4)
Physiotherapy at baseline	81/124 (67%)	45/67 (67%)
<b>Caregivers</b>		
Patient's partner	103/117 (88)	55/63 (87%)
Age (years)	67 (57.0–73.0)	65 (60.0–73.0)
Sex		
Men	37/117 (32%)	21/63 (33%)
Women	80/117 (68%)	42/63 (67%)
Educational level*		
High	45/117 (39%)	12 /62 (19%)
Middle	46 /117 (39%)	40/62 (65%)
Low	26/117 (22%)	10/62 (16%)
In paid employment	33/117 (28%)	19/62 (30%)

Data are median (IQR) or n/N (%). Some percentages do not sum up to 100% because of rounding. UPDRS III=Unified Parkinson's Disease Rating Scale, part III (score=108). MMSE=Mini-Mental State Examination (score=30). LED= Levodopa Equivalent Dose.\* Based on the Dutch educational system; low: primary education or low-level professional education; middle: secondary education or medium-level professional education; and high: tertiary education (bachelor degree or higher).† Data missing for one patient in the intervention group and one in the control group. ‡ Lower score suggests better functioning. † Data missing for two patients in the intervention group.

Subdomains of the Activity Card Sort were also assessed in a post-hoc analysis, and only one of four subdomains –instrumental activity participation– showed significant benefit in favour of the intervention group compared with the control group (mean difference 5.9%; 95% CI 1.8–10.0; p=0.006).

**Figure 3** Canadian Occupational Performance Measure scores

Estimated mean scores of the COPM-performance at 3 months (primary outcome) and 6 months in the intervention group (thick broken line) and in the control group (thick solid line). The estimated scores were calculated using a linear mixed model with adjustment for baseline values. The thin dashed lines show the 95% CIs. The vertical bars are observed means (dot, therapy; dash, control) and SEs.



**Table 2** Primary and secondary outcomes for patients

	Baseline		3 months		6 months		Difference between groups at 3 months		Difference between groups at 6 months	
	n	median(IQR)	n	median(IQR)	n	median(IQR)	mean (95% CI)	p value	mean (95% CI)	p value
<b>Canadian Occupational Performance Measure-performance scale (score 1–10)</b>										
intervention	124	4.3 (3.5–5.0)	122	5.8 (5.0–6.4)	120	5.7 (4.6–6.6)	1.2 (0.8 to 1.6)*	<0.0001	0.9 (0.5 to 1.3)	<0.0001
control	67	4.4 (3.8–5.0)	63	4.6 (3.8–5.5)	61	4.8 (4.0–5.5)	0.0 (ref)	..	0.0 (ref)	..
<b>Canadian Occupational Performance Measure-satisfaction scale (score 1–10)</b>										
intervention	124	4.2 (3.2–4.8)	122	5.6 (4.6–6.6)	120	5.7 (4.8–6.5)	1.1 (0.7 to 1.5)	<0.0001	0.9 (0.5 to 1.3)	<0.0001
control	67	4.3 (3.4–4.8)	63	4.6 (3.8–5.8)	61	4.8 (4.0–5.5)	0.0 (ref)	..	0.0 (ref)	..
<b>Perceive Recall Plan Perform system-phase 1 (%)</b>										
intervention	124	60.0 (40.0–75.0)	118	73.2 (50.0–88.9)	n/a	n/a	0.8 (-7.5 to 9.0)†	0.848	n/a	..
control	66	61.3 (33.3–80.0)	58	75.0 (50.0–92.3)	n/a	n/a	0.0 (ref)	..	..	..
<b>Activity Card Sort (%)</b>										
intervention	124	71.1 (57.2–82.7)	121	71.0 (56.9–83.5)	n/a	n/a	2.9 (-0.2 to 5.9)†	0.063	n/a	..
control	67	70.2 (61.5–79.2)	60	70.9 (56.5–81.8)	n/a	n/a	0.0 (ref)	..	..	..
<b>Utrecht Scale for Evaluation of Rehabilitation-Participation satisfaction scale (score 0–100)</b>										
intervention	123	60.0 (50.0–68.8)	122	58.3 (47.5–72.3)	120	55.9 (41.7–67.5)	3.2 (-0.6 to 6.8)	0.095	2.1 (-1.6 to 5.8)	0.262
control	66	61.1 (47.2–70.0)	62	59.2 (47.2–66.7)	61	57.5 (47.2–66.7)	0.0 (ref)	..	0.0 (ref)	..
<b>Parkinson's Disease Questionnaire (score 0–100)‡</b>										
intervention	122	35.5 (26.3–44.9)	118	34.5 (23.3–42.1)	119	36.3 (26.1–45.3)	-1.7 (-3.9 to 0.5)	0.135	-2.1 (-4.3 to 0.1)	0.056
control	65	34.6 (27.6–42.5)	60	33.5 (23.2–45.0)	60	35.6 (23.9–42.9)	0.0 (ref)	..	0.0 (ref)	..
<b>Euroqol 5 dimensions (score -0.33–1)</b>										
intervention	123	0.69 (0.65–0.78)	119	0.72 (0.57–0.81)	118	0.69 (0.57–0.81)	0.03 (-0.03 to 0.08)	0.351	0.02 (-0.03 to 0.07)	0.475
control	66	0.73 (0.57–0.81)	62	0.73 (0.57–0.81)	62	0.69 (0.57–0.78)	0.00 (ref)	..	0.00 (ref)	..
<b>Visual Analogue Scale for Quality of life (score 0–10)</b>										
intervention	124	7.0 (6.0–7.5)	121	7.0 (6.0–7.5)	120	6.0 (5.1–7.0)	0.3 (-0.1 to 0.6)	0.183	0.0 (-0.4 to 0.3)	0.822
control	66	7.0 (5.4–7.0)	61	7.0 (5.0–7.0)	61	7.0 (5.3–7.0)	0.0 (ref)	..	0.0 (ref)	..
<b>Fatigue Severity Scale (score 1–7)</b>										
intervention	124	5.0 (4.0–5.9)	122	5.0 (4.1–5.9)	120	5.1 (4.1–5.9)	0.1 (-0.2 to 0.4)	0.710	0.0 (-0.3 to 0.3)	0.846
control	66	4.9 (4.2–5.6)	62	4.8 (3.9–5.7)	61	4.9 (4.0–5.8)	0.0 (ref)	..	0.0 (ref)	..
<b>Becks Depression Inventory (score 1–63)‡</b>										
intervention	124	12.0 (8.0–18.0)	121	12.0 (7.0–16.0)	119	11.0 (7.0–17.0)	-1.4 (-3.0 to 0.3)	0.099	-0.8 (-2.5 to 0.8)	0.318
control	66	13.0 (9.0–17.0)	62	12.0 (8.3–18.9)	61	12.0 (9.0–17.8)	0.0 (ref)	..	0.0 (ref)	..
<b>Utrecht Proactive Coping Competence Scale (score 1–4)</b>										
intervention	124	2.7 (2.4–2.9)	120	2.7 (2.5–3.0)	117	2.7 (2.5–3.0)	0.1 (0.0 to 0.2)	0.101	0.1 (0.0 to 0.2)	0.266
control	65	2.6 (2.3–2.8)	62	2.6 (2.3–2.9)	61	2.6 (2.4–2.9)	0.0 (ref)	..	0.0 (ref)	..

All outcomes are secondary other than that marked with an asterisk. Group differences were estimated using linear mixed models for repeated data with adjustment for baseline values. For all measures unless otherwise stated, an increase in score over time suggests improvement. n/a=not applicable. Ref= reference value. \*Primary outcome † Absolute difference in percentage. ‡ Decrease in score over time suggests improvement.

**Table 3** Secondary outcome measures for caregivers

	Baseline		3 months		6 months		Difference between groups at 3 months		Differences between groups at 6 months	
	n	median (IQR)	n	median (IQR)	n	median (IQR)	mean (95% CI)	p value	mean (95% CI)	p value
<b>Zarit Burden Interview (score 0–88)*</b>										
intervention	117	18.0 (9.5–27.0)	114	18.0 (10.8–27.1)	112	19.0 (10.3–29.8)	-1.1 (-3.8 to 1.7)	0.440	-2.5 (-5.3 to 0.4)	0.089
control	62	18.5 (8.8–28.0)	59	22.0 (13.0–28.0)	53	24.0 (14.5–30.5)	0.0 (ref)	..	0.0 (ref)	..
<b>Zarit Burden Interview &gt; 20 at baseline*</b>										
intervention	52	29.5 (24.0–38.8)	50	27.1 (20.0–38.3)	48	29.5 (20.0–37.8)	-0.5 (-5.0 to 4.1)	0.835	-1.8 (-6.5 to 2.8)	0.438
control	27	29.0 (24.0–34.0)	28	26.5 (23.0–35.8)	26	29.5 (24.0–34.8)	0.0 (ref)	..	0.0 (ref)	..
<b>Zarit Burden Interview ≤20 at baseline*</b>										
intervention	65	10.0 (6.0–15.0)	64	11.0 (7.0–18.0)	64	12.0 (7.0–19.8)	-1.7 (-5.2 to 1.8)	0.334	-3.2 (-6.8 to 0.4)	0.082
control	35	9.0 (5.0–15.0)	31	14.0 (8.0–19.0)	27	17.0 (7.0–22.0)	0.0 (ref)	..	0.0 (ref)	..
<b>Visual Analogue Scale for Quality of life (score 0–10)</b>										
intervention	115	7.5 (7.0–8.0)	113	7.5 (7.0–8.0)	112	7.0 (7.0–8.0)	0.0 (-0.3 to 0.3)	0.819	0.2 (-0.1 to 0.6)	0.124
control	63	7.5 (7.0–8.0)	59	7.5 (7.0–8.0)	53	7.0 (6.3–8.0)	0.0 (ref)	..	0.0 (ref)	..
<b>Euroqol 5 dimensions (score -0.33–1)</b>										
intervention	115	0.84 (0.78–1.00)	112	0.84 (0.78–1.00)	104	0.84 (0.78–1.00)	0.06 (0.02 to 0.11)	0.006	0.04 (-0.01 to 0.09)	0.109
control	63	0.89 (0.78–1.00)	58	0.84 (0.78–1.00)	59	0.81 (0.78–1.00)	0.0 (ref)	..	0.0 (ref)	..
<b>Hospital Anxiety and Depression Scale-anxiety (score 0–21)*</b>										
intervention	117	5.8 (3.5–7.0)	112	4.7 (3.5–7.0)	111	5.8 (3.5–8.2)	-0.5 (-1.4 to 0.3)	0.209	-0.4 (-1.3 to 0.4)	0.296
control	63	4.7 (3.5–7.0)	59	4.7 (3.5–7.0)	53	4.7 (2.9–8.2)	0.0 (ref)	..	0.0 (ref)	..
<b>Hospital Anxiety and Depression Scale-depression (score 0–21)*</b>										
intervention	117	3.5 (1.2–5.8)	112	3.5 (1.2–5.8)	111	3.5 (1.2–5.8)	0.3 (-0.5 to 1.0)	0.529	0.0 (-0.9 to 0.8)	0.927
control	63	2.3 (1.2–4.7)	59	2.3 (1.2–4.7)	53	3.5 (1.2–5.8)	0.0 (ref)	..	0.0 (ref)	..
<b>Utrecht Proactive Coping Competence Scale (score 1–4)</b>										
intervention	114	2.8 (2.5–3.0)	109	2.9 (2.5–3.1)	109	2.9 (2.5–3.1)	0.0 (-0.1 to 0.1)	0.736	0.1 (0.0 to 0.2)	0.187
control	63	2.9 (2.5–3.1)	58	2.9 (2.5–3.2)	53	2.8 (2.5–3.0)	0.0 (ref)	..	0.0 (ref)	..
<b>Care minutes per day*</b>										
intervention	115	48.6 (7.2–104.4)	108	69.0 (10.2–166.2)	107	87.0 (12.6–201.6)	3.6 (-10.2 to 60.0)‡	0.758†	18.0 (-7.8 to 93.0)‡	0.537†
control	62	22.2 (4.2–129.6)	54	52.8 (3.6–121.2)	51	92.4 (8.4–213.0)	0.0 (-0.15 to 0.88)‡	..	9.0 (-4.8 to 120.0)‡	..

Group differences were estimated using a linear mixed model for repeated data with adjustment for baseline values. For all measures unless otherwise stated, an increase in score over time suggests improvement. Ref= reference value. †Decrease in score over time indicates improvement. ‡ Median(IQR)of the change compared with. † Mann Whitney test.

## Discussion

In this study, occupational therapy (the OTiP intervention) significantly improved patient's self-perceived performance in meaningful daily activities (primary outcome), had positive effects on satisfaction about performance of daily activities and on participation in instrumental activities, but did not improve caregiver outcomes, apart from the EuroQoL 5 dimensions scale at 3 months (panel).

At 3 months (immediately after the intervention), the group difference on self-perceived performance in meaningful activities (primary endpoint) was significant, and this persisted at 6 months. To clarify the clinical relevance of the efficacy of the intervention, we did a post-hoc analysis of the proportion of responders (defined as a clinically important change  $\geq 2$  points) in both groups. A significantly higher proportion of patients in the intervention group achieved a clinically relevant improvement on the COPM-P compared with those in the control group, but the proportion of responders in the intervention group was low. The estimated population effect of 1.2 was below the threshold for a clinically relevant change, presumably because the overall group included both responders and non-responders and a few patients whose performance score worsened. We used a conservative threshold of 2 points for a clinically relevant change, considering that a 2011 study on criterion responsiveness in outpatients found a lower optimum cut-off value of 1.4 for the COPM-P.<sup>42</sup> Using this cut-off, 62 (51%) of 122 patients in the intervention group achieved a clinically relevant improvement at 3 months, versus 11 (17%) of 63 in the control group. However, the results of this post-hoc analysis must be interpreted with caution. Work is needed to identify which factors related to the patient, environmental context or therapist might predict which patients are most likely to benefit from occupational therapy.

Qualitative analysis of our pilot study suggested that besides improved performance, occupational therapy affected many other factors related to daily functioning, such as increased insight and coping of patients and caregivers.<sup>12</sup> We therefore administered a battery of secondary measures. Satisfaction with performance in activities (COPM-S) showed a similar pattern as the COPM-P. This finding was expected, because perceived performance and satisfaction scores usually have a high correlation.<sup>33</sup> We also expected that improved perceived performance would lead to increased participation in activities (measured with the Activity Card Sort). However, this increase occurred only for one subscale (instrumental activities), but not for the overall score, nor for high-demand and low-demand leisure activities or social activities. Other secondary outcomes for patients, such as quality of life and coping, showed no effect. These scales might not be specific enough to detect the effects of the OTiP intervention.

Assessment of an individually tailored intervention is challenging because treatment aims are heterogeneous, hence the primary outcome measure should take into account differences between individuals in importance and relevance of daily

activities and perceived problems. We chose the COPM-P as the primary outcome because it best represents the nature of the OTiP intervention: it focuses on meaningful activities, allows for individual variation in priorities, and its rating is on the basis of person's own perceptions.<sup>29-31</sup> Moreover, the COPM is used commonly by occupational therapists as an instrument to identify and assess patients' goals.<sup>29</sup> However, using the COPM in a trial implies that priorities must be set during a baseline assessment with an assessor who will not undertake the intervention. For some patients, priorities might evolve over time on the basis of discussions and experiences.<sup>31,32</sup> Treatment goals covered in the intervention could be different from the priorities identified at baseline, as suggested by the relatively low mean congruence between COPM priorities at baseline and the actual intervention. This could have led to underestimation of the effect of the intervention because the COPM scored for the assessment only measures changes in the priorities that were set at baseline by the assessor; the effects of interventions addressing other goals were not captured.

The study protocol allowed for other interventions such as physiotherapy that might also improve activities. However, these additional interventions are unlikely to explain much of the benefits experienced by patients allocated to the OTiP intervention, because the number of patients who received physiotherapy was similar in both groups. Moreover, the number of patients receiving physiotherapy was similar between responders and non-responders in the intervention group. Another intervention with a potential positive effect on symptoms is PD drug treatment. However, because group differences in LED remained similar, a drug treatment bias is unlikely.

We found that the COPM difference between groups became smaller over time, which might suggest that a short period of occupational therapy results in temporary improvement, but that some form of maintenance therapy might be needed for sustained improvement. Further work is needed to study this possibility. By contrast, our expectation was that the COPM scores in the control group might improve over time, because COPM administration at baseline (which involves identification and prioritisation of affected activities) could increase the patients' awareness of their problems in daily activities, and prompt patients in the control group to seek solutions themselves. The reported data seem compatible with this theory.

The scarcity of effects for caregivers in the OTiP trial probably has various causes. First, inclusion was based on patient-specific criteria and the primary treatment focus was on patients' needs, so this design was possibly not suited to improvement of caregiver outcomes. In other multidisciplinary PD studies that involved caregivers as a secondary group, caregiver burden or anxiety actually increased.<sup>16,20</sup> Further evaluation of specific caregiver interventions or interventions focused on both patient and caregiver as a couple are warranted. Second, many caregivers experienced only low caregiver burden, probably because only few patients had advanced disease.

When caregiver burden is low, the motivation and scope for possible improvement is likely to be smaller, whereas the risk of inadvertently increasing caregiver burden is higher (because new treatment issues arise, or because of the time burden of attending treatment). However, this possible negative effect was not noted in our study.

A strength of the OTiP study was that the design and intervention were informed by the findings of a phase 2 feasibility study.<sup>43</sup> The intervention was delivered by experienced occupational therapists who were embedded in a structured multidisciplinary network, and who had received thorough baseline and follow-up training in treatment of PD patients according to practice guidelines. A further strength was the feasibility of the OTiP intervention: adherence to the OTiP-intervention protocol by therapists and satisfaction with the intervention among patients and caregivers were high.

This study had several limitations. The control group was not offered an intervention; hence, we cannot exclude that placebo effects contributed to the benefits experienced by patients in the intervention group. We chose this design for two reasons. First, because attention is an intrinsic part of occupational therapy, we thought that development of a matched placebo intervention would be difficult. Second, the design allowed us to assess what the added effect is of occupational therapy (including aspecific attention effects) over and above usual care, for which occupational therapy is rarely prescribed.<sup>19,20</sup> Further research is needed to disentangle the intrinsic effects of occupational therapy from the non-specific effects due to for example attention. Another shortcoming is that, because of low referral rates to occupational therapy, we needed to use a recruitment strategy that did not represent the referral processes in everyday clinical practice. We do not know what proportion of patients in the group who declined to be contacted for further information might have had an indication for occupational therapy, so we cannot decide whether the screened population is representative of the general PD population. Among the trial patients, many had mild disease, whereas in clinical practice most referrals to occupational therapy involve patients with more advanced disease. This finding could be explained by the eligibility criteria: patients who were eligible for the intervention, but who did not fit in the study design possibly represent the more advanced cases, because they were not living at home, or because they had severe comorbidity, or cognitive problems. Findings from a subanalysis of our results suggest that the COPM changes in the intervention group were similar across all disease stages, but this post-hoc analysis must be interpreted with caution. Nevertheless, our results suggest that mildly affected patients can also benefit from occupational therapy; this finding might change the referral process in practice. A final limitation is that we embedded the study within the context of usual PD care as it is currently organised in the Netherlands (ParkinsonNet concept). Thus we implied that all patients potentially had access to a structured healthcare environment with trained professionals from various disciplines

working in the community,<sup>23,25</sup> and our findings cannot be transferred automatically to other countries, unless an organisational context similar to the Dutch ParkinsonNet is implemented.

The results presented here focused on efficacy. Further analysis is needed to explore factors that are important for a successful intervention and implementation. Moreover, we will undertake analyses of cost-effectiveness. More research is needed to fully understand the effects of occupational therapy across the disease spectrum and the determinants affecting responsiveness.

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## Panel: Research in context

### Systematic review

We did a systematic search of intervention studies (reviews or trials) in Pubmed and CINAHL for studies that included the terms “occupational therapy” AND “Parkinson\*disease” and were published in English or Dutch between January 1, 1995 and October 1, 2013. We identified five systematic reviews of the effectiveness of occupational therapy,<sup>10,11,33-35</sup> and one meta analysis of occupational therapy related interventions.<sup>36</sup> The systematic reviews concluded that there is insufficient evidence for occupational therapy in Parkinson’s disease (PD) because of the scarcity of studies in this specialty. The few intervention trials included in the systematic reviews entailed group occupational therapy, which does not fully represent perceptions on client-centred occupational therapy. We found nine trials that assessed multidisciplinary interventions in PD including occupational therapy.<sup>15-17,20,37-41</sup> The efficacy of multidisciplinary care is inconclusive and what the contribution of occupational therapy was to the results of the trials cannot be established from the studies. Finally, we found two pilot intervention studies, including our own, which reported numerically positive effects of occupational therapy,<sup>12,13</sup> but these findings were not significant. We also found an additional occupational therapy pilot intervention study that showed a positive effect, but that study included patients with atypical parkinsonism (multiple system atrophy).<sup>14</sup>

### Interpretation

This is, to the best of our knowledge, the first large-scale randomised controlled trial to specifically assess the efficacy of occupational therapy in PD. The results of the present study suggest that occupational therapy – done according to guidelines and delivered in a Dutch multidisciplinary care context – improves self-perceived performance and satisfaction in daily activities, both immediately after the intervention (at 3 months) and after 6 months follow-up. The intervention did not have an effect on caregiver outcomes, apart from health-related quality of life at 3 months. Further process analysis might elucidate which factors are important for a successful intervention and implementation.

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### OTiP study Group

The OTiP study group consisted of all authors, G.F. Borm and E.M. Adang (contribution to research design; Radboud university medical center, Nijmegen), and the local coordinators in the regional hospitals: R Bruyn (Diakonessenhuis, Zeist ), T. Fennis (Ziekenhuis St. Jansdal, Harderwijk), J. Hoff (Sint Antonius Ziekenhuis, Nieuwegein/Utrecht), J. ten Holter (Deventer Ziekenhuis, Deventer), A. Hovestadt (Meander Medisch Centrum, Amersfoort), M. van Kesteren (Isala Klinieken, Zwolle), J.M.J. Krul and P.M. Laboyrie (Tergooiziekenhuizen, Hilversum and Bussum), F.E. Strijks (Gelre Ziekenhuizen, Zutphen), E. van Wensen (Gelre Ziekenhuizen, Apeldoorn).

### Acknowledgements

The study is funded by Prinses Beatrix Spierfonds and the Parkinson Vereniging, grant number WRO-029. We thank all patients and their primary caregivers for their participation and the occupational therapists who delivered the intervention: Corestha Warmenhoven (QuaRijn Kennis en Behandelcentrum, Doorn), Arianne Hilbers, Lisette Priem and Aletta Duynstée (Ergotherapie Praktijk Veluwe, Nijkerk), Ingrid Boekhoud (Advies en Behandelcentrum Careyn, Utrecht Stad), Carolien de Voogt (AxionContinu, Utrecht), Ellen Rozendal (Verpleeghuis de Hartkamp, Raalte), Maaïke van de Wetering-Wolswinkel (Verpleeghuis Norschoten, Barneveld), Birgit Wezelenburg, (Stichting Zorgpalet Baarn-Soest, Soest), Célie Journée (LEF Centrum, Zwolle and Ergotherapie La Vie, Broekland), Minka de Jong (Isala Klinieken, Zwolle), Jolanda Gons (Ergotherapie Gooi en Omstreken, Hilversum), Malou Overmeer (Vivium Zorggroep, Bussum), Carlijn Hansma (Ergotherapie Inovum, Loosdrecht), Astrid Rothuis (Praktijk voor Ergotherapie Dieren e.o., Dieren), Swanet Riphagen (Praktijk Ergotherapie Zutphen-Warnsveld, Warnsveld), Selma Sari and Rianne Merks (Zorggroep Apeldoorn en omstreken, Ergotherapie Thuis, regio Oost-Veluwe). We also like to thank Tiska Ikking and Soemitro Poerbodipoero (Amsterdam University of Applied Sciences), Esther Steultjens (HAN University of Applied Sciences) and all occupational therapy PRPP scorers for support in data collection. Finally, we thank Bart Kral for his assistance in administrative tasks.

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## Appendix

### Examples of occupational therapy interventions in Parkinson's disease

- I. An example of a meaningful activity in which a person with Parkinson's disease might experience limitations is grocery shopping. Occupational therapy aims at identifying the underlying causes, and the example below shows two possible causes (left side) as well as a set of tailored interventions that can be used in the context of this specific activity (right side):

Possible causes for impaired grocery shopping	Interventions				
<ol style="list-style-type: none"> <li>1. Planning an adequate shopping list (executive functioning deficits)</li> <li>2. Freezing in crowded situations (gait impairment, complexity environment)</li> </ol>	<ul style="list-style-type: none"> <li>• Training of cognitive strategies (planning and problem solving)</li> <li>• Restructuring daily routines to plan shopping during 'ON-moments', and during quiet times in the shop (simplifying activity context)</li> <li>• Application of cueing strategies (that are trained primarily in physical therapy) while walking in the shop</li> </ul>				
<ol style="list-style-type: none"> <li>II. The second example illustrates individual tailoring of the interventions to the abilities of the patient. The meaningful activity involves putting on a coat and the underlying problem is fastening the zipper due to impaired dexterity (specifically in situations of time pressure).</li> </ol>	<table border="1"> <thead> <tr> <th>Interventions for a patient with ability to learn strategies/methods</th> <th>Interventions for a patient with limited ability to learn new strategies/methods</th> </tr> </thead> <tbody> <tr> <td> <ul style="list-style-type: none"> <li>• Train application of time pressure management (cognitive strategy)</li> <li>• Advise sitting down when fastening a coat (minimise dual tasking)</li> <li>• Train focused attention to large finger/hand movements</li> </ul> </td> <td> <ul style="list-style-type: none"> <li>• Instruct the caregiver to avoid situations with time pressure (for the patient)</li> <li>• Instruct the caregiver to ask the patient to sit down</li> </ul> </td> </tr> </tbody> </table>	Interventions for a patient with ability to learn strategies/methods	Interventions for a patient with limited ability to learn new strategies/methods	<ul style="list-style-type: none"> <li>• Train application of time pressure management (cognitive strategy)</li> <li>• Advise sitting down when fastening a coat (minimise dual tasking)</li> <li>• Train focused attention to large finger/hand movements</li> </ul>	<ul style="list-style-type: none"> <li>• Instruct the caregiver to avoid situations with time pressure (for the patient)</li> <li>• Instruct the caregiver to ask the patient to sit down</li> </ul>
Interventions for a patient with ability to learn strategies/methods	Interventions for a patient with limited ability to learn new strategies/methods				
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## Chapter 5

### **Economic evaluation of occupational therapy in Parkinson's disease: a randomised controlled trial**



**Published as**

Sturkenboom IHWM, Hendriks JCM, Graff MJL, Adang EM, Munneke M, Nijhuis-van der Sanden MW, Bloem BR. Economic Evaluation of occupational therapy in Parkinson's disease: a randomized controlled trial. *Movement Disorders*, 2015; 30 (8): 1059-1067.

## Abstract

### Background

A large randomised clinical trial (the Occupational Therapy in Parkinson's disease [OTiP] study) recently demonstrated that home-based occupational therapy improves perceived performance in daily activities of persons with Parkinson's disease (PD). The aim of the present study was to evaluate the cost-effectiveness of this intervention.

### Methods

We performed an economic evaluation over a 6-month period for both arms of the OTiP study. Participants were 191 community-dwelling PD patients and 180 primary caregivers. The intervention group (n=124 patients) received 10 weeks of home-based occupational therapy; the control group (n=67 patients) received usual care (no occupational therapy). Costs were assessed from a societal perspective including healthcare use, absence from work, informal care, and intervention costs. Health utilities were evaluated using EuroQol-5d. We estimated cost differences and cost-utility using linear mixed models and presented the net monetary benefit at different values for willingness to pay per quality-adjusted life-year gained.

### Results

In our primary analysis, we excluded informal care hours because of substantial missing data for this item. The estimated mean total costs for the intervention group compared with controls were €125 lower for patients, €29 lower for caregivers, and €122 higher for patient-caregiver pairs (differences not significant). At a value of €40,000 per quality-adjusted life-year gained (reported threshold for PD), the net monetary benefit of the intervention per patient was €305 ( $p=0.74$ ), per caregiver €866 ( $p=0.01$ ) and per patient-caregiver pair €845 ( $p=0.24$ ).

### Conclusion

In conclusion, occupational therapy did not significantly impact on total costs compared with usual care. Positive cost-effectiveness of the intervention was only significant for caregivers.

## Introduction

Parkinson's disease (PD) is a complex and progressively disabling disease with an enormous impact on quality of life, for both the patient and the caregiver. PD also creates a high economic burden for the family and for society.<sup>1-3</sup> Although annual cost estimates vary between countries and studies, the general impression is that costs rise with disease progression.<sup>2,4</sup> The main direct cost drivers in PD are institutionalisation and medication.<sup>1,2,4</sup> In addition, with disease progression, nonmedical costs because of productivity loss or informal care also rise.<sup>3</sup> Since PD has no cure, employing effective strategies to optimise daily functioning and social participation of patients, and to enable caregivers to uphold a supportive role, is important. Moreover, limited health and social care budgets necessitate these strategies to be cost-effective. Occupational therapy is a relatively low-cost allied health intervention specifically focusing on optimizing daily functioning and participation (figure 1).<sup>5,6</sup> Recently, a large-scale randomised controlled trial of Occupational Therapy in PD (the OTiP study), demonstrated that a 10-week home-based occupational therapy intervention improved patient's perceived performance and satisfaction in daily activities at 3 months and 6 months after baseline.<sup>7</sup> The intervention also positively influenced quality of life of caregivers. However the cost-effectiveness has not yet been established, and this is the purpose of this paper.

Only a few economic evaluations within the field of allied healthcare in PD have been conducted,<sup>8-10</sup> but none addressed cost-effectiveness of occupational therapy in PD. Because occupational therapy addresses performance in complex daily activities, our hypothesis was that occupational therapy might reduce costs by alleviating the need for homecare or informal support or necessity for hospitalisation. Additionally, occupational therapy addresses the caregiver's needs in supporting the patient, and this might reduce costs such as informal care and caregiver's absence from work. Conversely, we expected costs for aids and adaptations to increase, because advice on environmental modifications can be a specific occupational therapy intervention strategy. Moreover, occupational therapists might signal the need for involvement of other healthcare professionals, and thus we anticipated costs for consultations of other disciplines to increase. Overall, we hypothesized that the benefits would outweigh the extra investments, and that a 10-week occupational therapy intervention would save costs over 6 months.



## Methods

We conducted an economic evaluation from a societal perspective over a 6-month period in parallel with a multicentre, assessor-masked, randomised controlled clinical two-arm efficacy trial within the context of specialised networks for PD (ParkinsonNet).<sup>7,11</sup> Note that the study was powered on the primary endpoint of the efficacy study, namely, perceived performance in daily activities, and not on cost-effectiveness. Ethical clearance was provided by the medical ethical committee of Arnhem-Nijmegen (NL27905.091.09/ABR27905).

### Participants

As reported elsewhere,<sup>7</sup> 191 PD patients and 180 primary caregivers from 10 hospitals (in 9 ParkinsonNet regions) were included in the OTiP study between April 2011 and November 2012. Patients fitted the inclusion criteria of being diagnosed with PD according to the UK Brain Bank criteria,<sup>12</sup> living at home, and reporting difficulties in daily activities. Patients with atypical parkinsonism, those who had received occupational therapy in the preceding 3 months, had predominant disabling co-morbidity, had insufficient understanding of Dutch language or a Mini Mental State Examination score less than 24 had been excluded. A primary caregiver could participate when willing and available. Patients and caregiver provided written informed consent before voluntary participation.

After baseline assessment, participants were stratified by region, and randomly assigned to the experimental or control group in a ratio of 2:1 using a computer-generated minimization algorithm. Minimization factors were PD severity indexed by Hoehn and Yahr score less than 3 versus 3 or higher, baseline perceived performance in daily activities measured with Canadian Occupational Performance Measure (COPM) score <5 vs. ≥5, gender, age (<65 vs. ≥65), and receiving physiotherapy at baseline yes versus no.

### Intervention

In brief, patients and their caregivers in the intervention group received 10 weeks (maximum 16 h) of individualised therapy according to the Dutch guidelines of occupational therapy in PD within the first 3 months after baseline assessment.<sup>5,7</sup> The intervention was delivered by 18 trained occupational therapists in the patient's home environment and focused on improving performance in daily activities selected and prioritised by the patient. Caregiver's needs in supporting the patient in daily activities were evaluated and addressed if required. The control group was not allowed to receive occupational therapy. Both groups could receive all other medical, psychosocial or allied healthcare interventions as usual. More details are reported elsewhere.<sup>7,11</sup>

### Occupational therapy in Parkinson's disease

**Aim:** enhancing daily activity performance and facilitating engagement in activities (self-care, productivity, leisure) at home or in the community

**Overarching principles:** client centred, individually tailored, embedded in the performance context

**Interventions:** targeting person, activities and/or environmental context

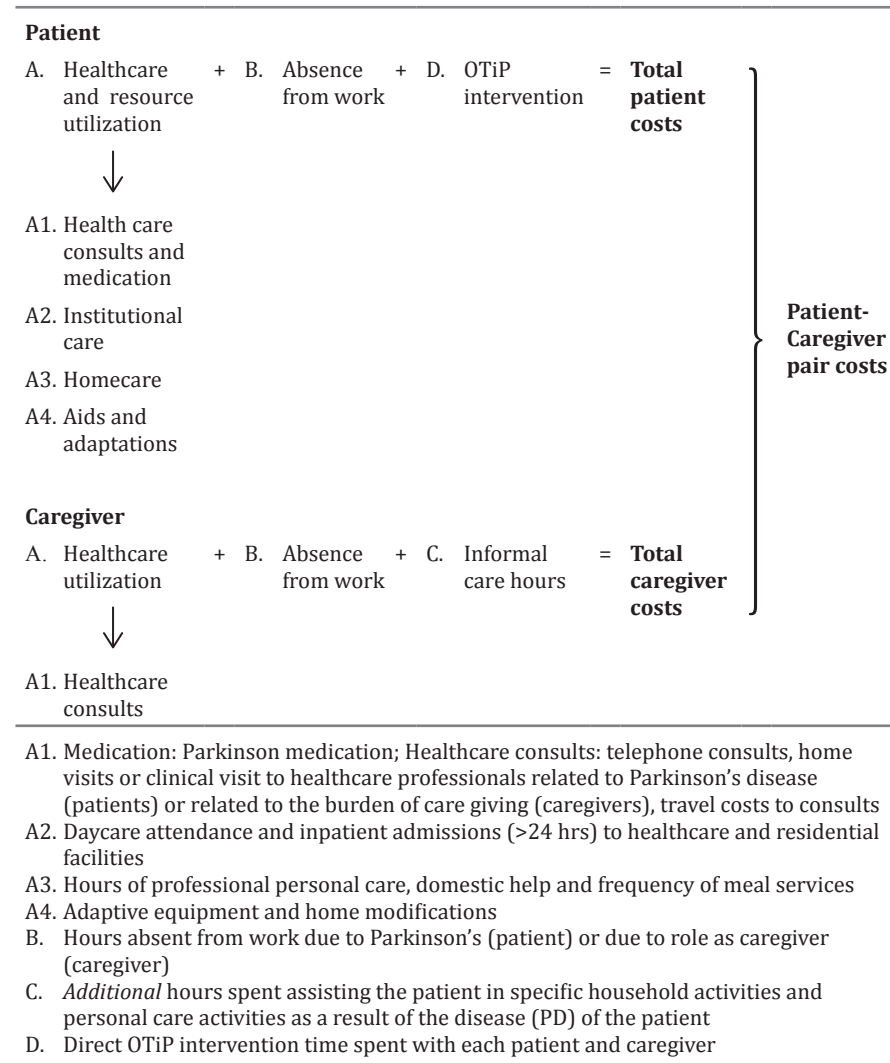
- Person: coaching to increase insight and self efficacy, training of performance strategies
- Activities: simplification of tasks, optimizing daily routines
- Physical environment: advise on appropriate aids and home modifications
- Social environment: coaching caregiver and training skills in supporting the patient

**Figure 1** Occupational Therapy in Parkinson's disease

### Assessment of costs

To enable cost analysis from a societal perspective, we collected cost data for the following categories: A) healthcare and resource utilisation, B) absence from work, C) informal care hours, and D) OTiP intervention (Figure 2). At baseline, 3 months and 6 months, the patients filled in questionnaires retrospectively for the previous 3 months. Similarly, caregivers filled in questionnaires on consultations of healthcare professionals for issues related to the burden of care. At baseline, the questions on hours of informal care provision were filled in through structured interview with the assessor. At 3 months and 6 months, the caregivers filled in these questions themselves. Occupational therapists used a time sheet in the patients' records to register the number of hours spent with each patient and caregiver.

Costs (in euro) per variable were calculated by multiplying volumes for 3 months with the unit cost prices (see supplementary information, Appendix 1). These unit cost prices were based on standard prices as stated in the Dutch manual for costing research in healthcare<sup>13</sup> or the Healthcare Insurance Board reference database.<sup>14</sup> We used a standard transportation cost price for each clinical healthcare visit. Cost prices for medication were obtained from a formal Dutch reference database for medication.<sup>15</sup> We valued adaptive equipment (per type) using the average market rate from a Dutch online database for adaptive equipment<sup>16</sup> and divided this rate by the number of trimesters in which the product would economically be written off as the cost per trimester. A similar approach of using trimester cost price was used for investment costs of other products or home adaptations. Absence from work of patients and caregivers was calculated according to the friction cost method,<sup>13</sup> using the same standard for full time hours (1,540 per year) for both employed and self-employed participants. When a person had no paid work, costs for absence were set at 0. According to the Dutch manual for costing research, we valued informal care



**Figure 2** Positioning of total costs per patient, caregiver and patient-caregiver pair by their specific cost categories

at a standardised hourly rate for a domestic cleaner.<sup>13</sup> When a patient participated without a caregiver and informal practical support was not present, the costs for the caregiver were all set at zero.

### Assessment of health utilities

Patients and caregivers each filled in the Euroqol-5 dimensions scale (EQ-5d),<sup>17</sup> a generic five-item validated health-related quality of life questionnaire. It allows a standardised approach for obtaining health utilities for use in QALY calculations and it is recommended for use in cost-utility studies in PD.<sup>18</sup>

### Statistical methods

If a patient or caregiver had indicated using a certain resources (e.g., speech therapy) but had not reported frequency, the mean frequency of all participants in the respective study arm using that resource was used as value. The calculations with respect to “informal care hours” were performed if at least 66% of its associated items were non-missing (i.e., had valid values). We aggregated the costs reported at 3 and 6 months after baseline to get costs for the 6-month period. We summed costs within each cost category and subsequently calculated total cost variables for each patient, caregiver and patient-caregiver pair. Utility was calculated as quality-adjusted life-year (QALY) over a 6-month timeframe using the trapezium rule. QALYs for patients and caregivers are derived using the EQ-5d health tariffs for the Dutch population (utility score -0.33 to 1.0).<sup>19</sup>

To study between-groups differences for costs and QALYs, we used linear mixed models with dependent variable cost or QALY. The independent fixed variables were group (control, OTiP intervention), baseline cost, and the minimization factors. Region was treated as a random variable. The model is specified in detail in the supplementary information, Appendix 2. The analyses were performed following the intention-to-treat principle. We present the observed costs (median range) for baseline and for the 6-month period and the baseline-adjusted mean difference between groups over 6 months with 95% confidence intervals.

Second, the net monetary benefit (NMB) statistic was used to evaluate cost-effectiveness.<sup>20</sup> The formula is:  $NMB = \text{Willingness To Pay} * \Delta QALY - \Delta \text{Costs}$ . The outcome indicates the monetary gains or costs of an intervention at explicit willingness to pay (WTP) thresholds. When the NMB (and the 95% lower-level confidence interval [CI]) is higher than 0, the experimental intervention is considered significantly cost-effective. Five WTP threshold for a QALY gained were used: 0, 20,000, 40,000, 60,000 and 80,000 euro. In the Netherlands, the illness burden of PD is 0.497 (scale 0-1),<sup>21</sup> and this corresponds to a WTP per QALY of nearly €40,000.<sup>22</sup> Therefore we used the 40,000 WTP threshold as reference value. Again, to study the differences between groups in NMB, a similar linear mixed model was used, but with NMB as dependent variable.

## Results

Table 1 presents the baseline characteristics relevant for the economic evaluation. Of the 191 included patients, nine were lost to follow-up at 6 months (intervention n=3; control n=6); for caregivers this was 14 (intervention n=4; control n=10). After handling missing data, a considerable number of incomplete cases were found in total costs of caregivers and patient-caregiver pairs, mainly because of missing data in the cost category “informal care hours” (intervention: 47 of 124, control: 29 of 67). Therefore, total costs and cost-effectiveness were analyzed both with and without this cost category. We considered the analyses without “informal care hours” as our primary analysis.

**Table 1** Baseline characteristics patients and caregivers

	Intervention group	Control group
<b>Patients</b>		
N	124	67
Age (years)	71.0 (63.3–76.0)	70.0 (63.0–75.0)
Men	78 (63%)	41 (61%)
In paid employment*	16 (13%)	12 (18%)
Disease duration (years) †	6.0 (4.0–10.0)	6.0 (3.0–11.0)
Hoehn and Yahr stage ‡		
1	31 (25%)	15 (22%)
2	46 (37%)	32 (48%)
3	44 (36%)	16 (24%)
4 or 5	3 (2%)	4 (6%)
<b>Caregivers</b>		
N	117	63
Partner-relationship to patient	103 (88%)	55 (87%)
Age (years)	67 (57.0–73.0)	65 (60.0–73.0)
Men	37 (32%)	21 (33%)
In paid employment	33 (28%)	19 (30%)

Data are median (IQR) or n (%). Some percentages do not sum up to 100% because of rounding.

\*Data missing for one patient in control group. †Data missing for one patient in the intervention group and one in the control group. ‡Lower score suggests better functioning.

Baseline data previously published in Sturkenboom et al.<sup>7</sup>

## Cost differences

The observed costs and estimated mean cost differences between groups for cost categories and total costs are presented in Table 2. In the primary analysis (i.e. excluding “informal care hours”), the mean total costs for the intervention group compared with controls were €125 lower for patients, €29 lower for caregivers and €122 higher for patient-caregiver pairs (all differences non-significant). When including “informal care hours” in the analysis, estimated mean costs were non-significantly higher in the intervention group for caregivers (€204) and for patient-caregiver pairs (€1,219).

Results for cost differences in separate cost categories showed that in the category “healthcare and resource use”, only the costs for institutional care of the patient were significantly lower in the intervention group (€1,458 euro; p=0.04). Costs for “absence from work” were €282 lower for patients of the intervention group (not significant). The provision of informal care incurred non-significantly higher costs (€215) in the intervention group. The mean difference in OTiP intervention costs (only utilised in the intervention group) was €760.

## Monetary benefits

At 6 months, the estimated differences between groups in utility scores (EQ-5d) of patients, caregivers and patient-caregiver pairs did not reach level of significance but were in favor of the intervention group (Table 2). The mean net monetary benefit of the intervention at a WTP value of €40,000 is per patient €305 (95% CI -1,538 to 2,148; p=0.74), per caregiver €866 (95% CI; 223 to 1,509; p=0.01) and per patient-caregiver pair €845 (95% CI -1,228 to 2,917; p=0.24) (Figure 3). When society is willing to pay €20,000 per QALY gained the net benefit of the intervention for the caregiver is positive, with a probability of 95%.

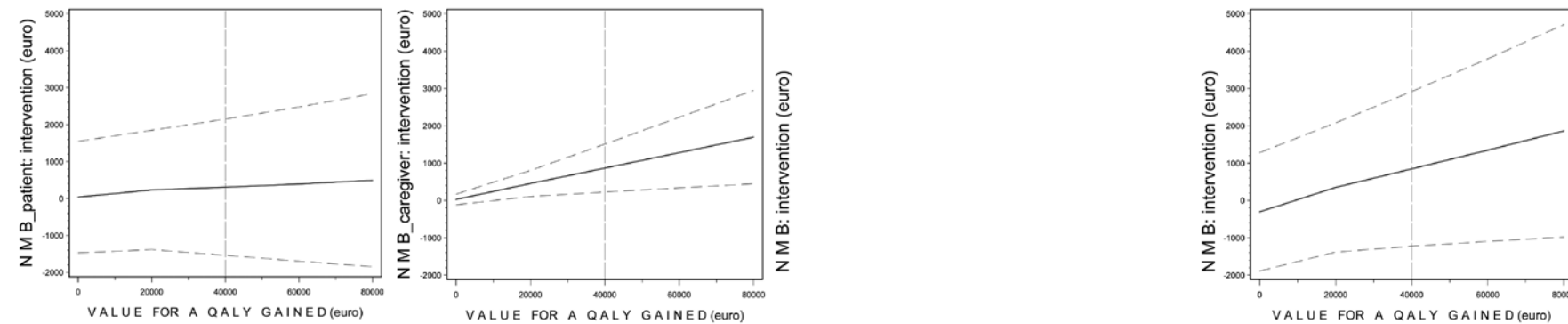
**Table 2** Group differences in costs and health utilities estimated using a linear mixed model with adjustment for baseline values and minimization factors

	Baseline observed			6 months observed			Estimated difference between groups over 6 months
	n	n <sub>u</sub>	Median (min–max)	n	n <sub>u</sub>	Median (min–max)	Mean (95% CI)
<b>Costs patients</b>							
Healthcare consults and medication							
intervention	124	123	860 (0–21,117)	118	118	1940 (261–41,285)	75 (-167 to 317)
control	66	65	938 (0–11,104)	61	60	1883 (0–22,006)	ref
Institutional care							
intervention	124	14	0 (0–9,935)	120	21	0 (0–18,568)	-1458 (-2,825 to -91)*
control	66	9	0 (0–4,258)	61	16	0 (0–46,430)	ref
Aids and adaptations							
intervention	124	29	0 (0–328)	122	66	7 (0–999)	14 (-18 to 47)
control	66	12	0 (0–294)	62	22	0 (0–588)	ref
Homecare							
intervention	124	43	0 (0–9,681)	120	21	0 (0–21,992)	83 (-615 to 782)
control	66	20	0 (0–9,586)	60	54	0 (0–23,308)	ref
Absence work							
intervention	124	4	0 (0–11,966)	122	2	0 (0–23,932)	-282 (-913 to 349)
control	66	3	0 (0–3,357)	61	5	0 (0–15,074)	ref
OTiP intervention							
intervention	124	0	0 (0–0)	123	123	749 (327–1,635)	760 (714 to 806)**
control	67	0	0 (0–0)	67	0	0 (0–0)	ref
Total costs patient							
intervention	124	124	1302 (53–21,591)	117	117	3448 (832–43,622)	-125 (-1,651 to 1,401)
control	66	66	1274 (202–11,683)	60	60	2372 (152–36,613)	ref
<b>Costs caregivers</b>							
Healthcare consults							
intervention	118	51	0 (0–1,306)	113	60	35 (0–2,717)	-32 (-171 to 107)
control	63	33	30 (0–1,225)	54	34	53 (0–1,536)	ref
Absence work							
intervention	119	4	0 (0–622)	113	7	0 (0–373)	4 (-15 to 22)
control	64	5	0 (0–466)	54	7	0 (0–249)	ref
Informal care hours (IC)							
intervention	112	96	903 (0–9,196)	77	65	2543 (0–14,046)	215 (-1,033 to 1,434)
control	61	48	491 (0–7,780)	38	34	3196 (0–12,359)	ref
Total costs caregiver excluding IC							
intervention	118	51	0 (0–1,306)	113	64	81 (0–2,717)	-29 (-172 to 114)
control	63	37	35 (0–1,225)	54	37	75 (0–1,536)	ref
Total costs caregiver including IC							
intervention	112	98	1087 (0–9,196)	77	69	2,923 (0–12,592)	204 (-1,059 to 1,467)
control	60	50	736 (0–9,462)	38	35	3294 (0–12,359)	ref

**Table 2** Continued

	Baseline observed			6 months observed			Estimated difference between groups over 6 months
	n	n <sub>u</sub>	Median (min-max)	n	n <sub>u</sub>	Median (min-max)	Mean (95% CI)
<b>Costs patient-caregiver pairs</b>							
Total costs patient + caregiver excluding IC							
intervention	118	118	1,400 (53-21,641)	111	111	3874 (832-44,436)	122 (-1,483 to 1,727)
control	63	63	1,395 (202-12,849)	54	54	2726 (256- 37,656)	ref
Total costs patient + caregiver including IC							
intervention	112	112	2,604 (201-22,884)	77	77	6412 (1,055-54,927)	1219 (-1,248 to 3,739)
control	60	60	2,452 (296-20,458)	38	38	5247 (845- 26,658)	ref
<b>Health utilities</b>							
Patients EQ-5d							
intervention	123	n/a	0.69 (-0.11-1.00)	118	n/a	0.69 (-0.11-1.00)	0.02 (-0.03 to 0.07)
control	66	n/a	0.73 (-0.13-1.00)	62	n/a	0.69 (-0.13-1.00)	ref
Caregivers EQ-5d							
intervention	115	n/a	0.84 (0.22-1.00)	104	n/a	0.84 (0.26-1.00)	0.04 (-0.01 to 0.09)
control	63	n/a	0.89 (0.17-1.00)	59	n/a	0.81 (0.25-1.00)	ref
Patient-caregiver pairs EQ-5d							
intervention	115	n/a	1.54 (0.44-2.00)	110	n/a	1.57 (0.39-2.00)	0.05 (-0.03 to 0.13)
control	63	n/a	1.57 (0.65-2.00)	51	n/a	1.57 (0.52-2.00)	ref

Costs are presented in euro (1 EUR ≈ 1.30 USD; conversion rate Sept 15<sup>th</sup> 2014). Observed baseline data cover a period of preceding 3 months; observed 6 months data cover a period of preceding 6 months, n<sub>u</sub> = number of health service users (i.e. costs>0) of each cost component, IC= informal care hours. Observed health utilities present EQ5d (EuroQol-5d) utility scores at baseline and at 6 months. n/a = not applicable  
 \* p value <0.05; \*\* p value<0.0001; ref=reference value.



**Figure 3** The net monetary benefit in the intervention group compared with the control group against the value for a QALY gained for patients (left), caregivers (middle) and patient- caregiver pairs (right)

Solid line= net monetary benefit. Dashed lines = confidence intervals

## Discussion

Previously, we established efficacy of the OTiP intervention for improving patient's self-perceived performance in daily activities, as compared with usual care.<sup>7</sup> The current cost-effectiveness study over a 6-month time horizon demonstrated no significant differences in total costs between both treatment arms, but a significant and substantial saving on institutional care in the intervention group. Moreover, compared with usual care no net monetary benefit of occupational therapy for patient-caregiver pairs was found.

### Cost-differences

The hypothesis that costs for healthcare consults might be higher in the intervention group, because occupational therapists might signal the need for input from other professionals, was not confirmed.

Moreover, we expected a lower need for institutional care of the patient because occupational therapy supports adaptations in daily functioning. Our results support this hypothesis, because mean total costs for institutional care over 6 months were significantly and substantially lower (€1,458) than in the control group. This is highly relevant because (Dutch) healthcare policies are directed at supporting patients to live at home for as long as possible to save on institutional care. Our finding is in line with other evaluations of Parkinson care in the Netherlands.<sup>9,23</sup> The ParkinsonNet concept (a series of specialised multidisciplinary networks for PD) proved to save on institutional care.<sup>9,23</sup> In our study, both groups were embedded within ParkinsonNet, and involvement of occupational therapy was the only contrast. Therefore, our OTiP results suggest that occupational therapy can reduce cost for institutional care on top of the savings through ParkinsonNet care. In contrast to our hypothesis, the costs for adaptive equipment and home modifications were similar between groups. An additional descriptive analysis indicated that persons in the intervention group did report more items over 6 months (88 in the intervention group; 14 in the control group). Moreover, items reported covered a greater variety, such as various types of mobility support (e.g., bed canes, wheeled walkers) and many small items for specific activities (e.g. cutlery, playing card holder, medication alarm). This indicates a different pattern in utilisation of assistive devices and home modifications between groups. A possible explanation for similar costs in both groups might be that the method to spread costs over economic lifetime prevented finding great differences in costs over a 6-month time horizon.

Regarding absence from work, only a small proportion of patients in the study were still in paid work (13% intervention group; 18% control group), and therefore a significant difference between groups was not to be expected. To test a hypothesis related to productivity loss, one should conduct a study specifically in the subgroup

of young-onset PD patients and evaluate not only absenteeism but also PD-related presenteeism.

For “informal care hours”, we had expected that the focus on both patient's and caregiver's issues in the intervention would result in reduced hours of care provision, as was found in another Dutch study on home-based occupational therapy in the field of dementia.<sup>24</sup> Additionally, in a study focusing on enhancing PD patients' mobility (by physiotherapy embedded in ParkinsonNet), an indirect effect of reduction in informal care hours was demonstrated in the intervention group.<sup>9</sup> By contrast, here we found higher informal care costs for the intervention group, but this difference was not statistically significant. Yet, we have to interpret these results with caution, because the substantial missing data in the domain of informal care hours can have led to selection bias. Reliable and complete measurement of informal care provided as a consequence of disease is difficult, because supporting the patient in doing tasks such as cooking a meal or visiting friends are also part of normal social interaction in a family or partner relationship.<sup>25-27</sup> We had thought to partly tackle this measurement issue by letting assessors administer the questionnaire at baseline by interview during which the instruction “additional hours due to...” could be stressed. However, in hindsight, this approach did not appear to be able to prevent a high number of missing data and bias at follow-up.

### Monetary benefits

The small and non-significant incremental net monetary benefit for patients, might partly be explained by a poor responsiveness of the EQ-5d as an effect measure in PD, which has been highlighted in the literature.<sup>28,29</sup> In contrast, a more sensitive measure for effectiveness of the OTiP intervention was a specific measure on perceived daily activity performance (COPM).<sup>7,30</sup> However, policy makers need to select across interventions and diseases and the EQ-5d has the benefit of being a general measure with referenced WTP thresholds. Therefore it is still the recommended measure for economic evaluations in PD.<sup>18</sup> The OTiP study was, however, not powered on the cost-effectiveness outcomes (i.e., costs, EQ-5d) but rather on the COPM.

### Strengths and limitations

The strength of the OTiP study is that we used a randomised controlled design and took a societal perspective for the economic analysis. We acquired data on a wide range of cost categories, covering all possible areas the intervention might impact on. This allowed us to get insight into total cost differences, but also provided opportunity to reflect on the different results for separate cost categories.

Because cost data are calculated from a large number of items, the cost variables are obviously subject to incomplete data. To handle this, we followed standard procedures and made logical interpretations. To study the sensitivity of these inter-

pretations on the conclusions we also analyzed the data without replacing missing. We found similar results for patients, caregivers and patient–caregiver pairs, and, as expected, with some wider confidence intervals. We concluded that the results presented are invariant regarding these handling rules.

An imperative limitation of our study was that we collected the resource utilisation data through means of retrospective self-report questionnaires. This method is commonly used but prone to recall bias and high rates of missing data,<sup>31</sup> especially in PD patients who have high healthcare consumption. In the field of economic evaluation the need for consensus on best tools and methods for accurate recourse-use measurement has been highlighted.<sup>31</sup> With regard to caregivers, we already discussed the issues of reliably measuring informal care hours.

Another limitation was the relatively short period of follow-up for an economic evaluation. The period was only extended to 6 months' follow up (i.e., 3 months after the intervention was finished). In the efficacy study, we found that the differences between groups became smaller from 3 to 6 months' assessment, indicating limited sustainability of the effect and need for some form of maintenance therapy. This would require additional intervention costs and the cost-effectiveness of that approach would need to be evaluated.

A final limitation is that the outcomes are linked to the societal and healthcare context at the time of study, because this context influences availability and accessibility of resources.<sup>32</sup> In the Netherlands basic healthcare insurance is obligatory, and in care planning, the professionals and patient take into consideration what resources are covered in the basic health insurance or other legislations and what will costs be for the individual. The exact consequences of these considerations for resource use are not known, and we have not differentiated between cost covered (societal costs) and individual costs in this study.

### Conclusion and future perspectives

This study provides initial insights into the cost-effectiveness of a home-based occupational therapy intervention for PD patients in the Netherlands. This intervention (proved to be effective in a recently published efficacy study) did not significantly impact on total costs compared with usual care over a 6-month period. Cost-effectiveness was significantly better for caregivers allocated to occupational therapy, but other cost-effectiveness analyses were comparable for occupational therapy and usual care. We are performing a process evaluation of the study evaluating what factors are important for a successful occupational therapy intervention. This may provide information on ways to optimise efficiency of the intervention for patients, caregivers and pairs. Future research focusing on longer term evaluations of costs and effects is warranted, but considering the findings from the efficacy study, the design should allow intermittent follow up. The study highlighted the challenge of

efficient and reliable measurement of costs when conducting economic evaluations from a societal perspective. To measure informal care hours we recommend further exploration of the concept of informal care giving in a degenerative disease such as PD: How do caregivers perceive differentiation in support provided as a consequence of PD versus usual family support, and what is the relationship with quality of life?

### Acknowledgements

The study was financially supported by Prinses Beatrix Spierfonds and Parkinson Vereniging; grant number WRO-029.

We thank all participants (patients and caregivers), and the partaking occupational therapists for their contribution to the study: Coestha Warmenhoven (QuaRijn Kennis en Behandelcentrum, Doorn), Arianne Hilbers, Lisette Priem and Aletta Duynstée (Ergotherapie Praktijk Veluwe, Nijkerk), Ingrid Boekhoud (Advies en Behandelcentrum Careyn, Utrecht Stad), Carolien de Voogt (AxionContinu, Utrecht), Ellen Rozendal (Verpleeghuis de Hartkamp, Raalte), Maaïke van de Wetering-Wolswinkel (Verpleeghuis Norschoten, Barneveld), Birgit Wezelenburg, (Stichting Zorgpalet Baarn-Soest, Soest), Célie Journée (LEF Centrum, Zwolle and Ergotherapie La Vie, Broekland), Minka de Jong (Isala Klinieken, Zwolle), Jolanda Gons (Ergotherapie Gooien Omstreken, Hilversum), Malou Overmeer (Vivium Zorggroep, Bussum), Carlijn Hansma (Ergotherapie Inovum, Loosdrecht), Astrid Rothuis (Praktijk voor Ergotherapie Dieren e.o., Dieren), Swanet Riphagen (Praktijk Ergotherapie Zutphen-Warnsveld, Warnsveld), Selma Sari and Rianne Merks (Zorggroep Apeldoorn en omstreken, Ergotherapie Thuis, regio Oost-Veluwe). We thank the local coordinators in the regional hospitals: R Bruyn (Diakonessenhuis, Zeist), T. Fennis (Ziekenhuis St. Jansdal, Harderwijk), J. Hoff (Sint Antonius Ziekenhuis, Nieuwegein/Utrecht), J. ten Holter (Deventer Ziekenhuis, Deventer), A. Hovestadt (Meander Medisch Centrum, Amersfoort), M. van Kesteren (Isala Klinieken, Zwolle), J.M.J. Krul and P.M. Laboyrie (Tergooiziekenhuizen, Hilversum and Bussum), F.E. Strijks (Gelre Ziekenhuizen, Zutphen), E. van Wensen (Gelre Ziekenhuizen, Apeldoorn).

We thank Tiska Ikking and Soemitro Poerbodipoero (Amsterdam University of Applied Sciences) for support in data collection and Yvonne Veenhuizen (Radboudumc, Nijmegen) for her support in both data collection and data cleaning. We also thank Bart Kral for his secretarial assistance.

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## Appendix 1 Price per unit for the cost variables

Cost categories	Cost variables	Unit of measurement	Unit Cost (€)	Source
<b>A. Healthcare and resource utilisation</b>				
A1. Healthcare consults and medication	General practitioner	Consult <sup>1,2</sup>	28.99	a
	Neurologist	Consult <sup>1,2</sup>	74.54	a
	Parkinson nurse specialist	Consult <sup>3</sup>	15.79	a
	Physiotherapy	Consult <sup>2</sup>	35.30	b
	Speech therapy	Consult <sup>2</sup>	31.78	b
	Social work	Consult <sup>1,2</sup>	67.29	a
	Psychologist	Consult <sup>1</sup>	82.82	a
	Psychiatrist	Consult <sup>1</sup>	106.63	a
	Travel costs healthcare consults	Per consult	6.00	c
	Parkinson medication	Dose	Depends on type	c
A2. Institutional care	Day treatment	Day	97.36	a
	Hospital admission	Night	473.11	a
	Residential care	Night	90.16	a
	Nursing home	Night	246.39	a
	Rehabilitation Centre	Night	351.99	a
A3. Aids and adaptations	Aids and adaptations	Type	Purchasing price/depreciation time in trimesters	d
A4. Homecare	Personal care assistance	Hour	45.35	a
	Domestic care assistance	Hour	12.94	a
	Home-delivered meals	Meal	6.22	e
	Meal in residential setting	Meal	6.93	e
	Other costs (i.e., paid support)		As reported	f
B. Absence from work	Absence from work	Hour	31.08	a
C. Informal care hours	Caregiver time	Hour	12.94	a
D. Intervention	Occupational therapy	Home visit per hour	81.76	b

Prices are indicated in euro. (1 EUR ≈ 1.30 USD; conversion rate Sept 15<sup>th</sup> 2014).

<sup>1</sup>Telephone consult = consult x 0.25; <sup>2</sup> Home visit = consult x 1.5 <sup>3</sup>telephone consult= ½ hr consult x0.5 and home visit= ¼ hr consult x3.

a Reference prices in Dutch Manual for Costing: Methods and Reference Prices for Economic Evaluations in Healthcare, indexed to 2011.<sup>1</sup>

b Reference database of the Dutch Health Care Insurance Board<sup>2</sup> (prices 2011)

c Fixed price 7 km one-way\*0,20 per km+ 3,00 parking.

c Dutch Pharmacotherapeutical Kompas<sup>3</sup>

d for purchasing price: Dutch reference database for adaptive equipment (mean price)<sup>4</sup>

e Average market price different providers

f Patient reported

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## Appendix 2 Specifications Linear Mixed Model

The initial model we used is as follows:  $Y_{ij}(t_g) = \beta_0 + \beta_1 Y_i(t_0) + \beta_2 E_i + \beta_3 H_i + \beta_4 C_i + \beta_5 G_i + \beta_6 A_i + \beta_7 P_i b_j + \epsilon_i$ . Where  $i$  refers to subject and  $j$  to the region, with the fixed effects  $\beta$  and the random effect  $b$  related to region.  $E$ ,  $H$ ,  $C$ ,  $G$ ,  $A$ ,  $P$  and are indicator variables for OTiP group, Hoehn and Yahr score  $\geq 3$ , COPM score  $\geq 5$ , gender male, age  $\geq 65$  years, physiotherapy at baseline, respectively, and  $\epsilon_i$  is the normal distributed residual with mean zero. The regression parameters with standard error were used to estimate the effect of the OTiP intervention compared with the control, with the appropriate 95% confidence interval.

## Chapter 6

### **Process evaluation of a home-based occupational therapy intervention for Parkinson's patients and their caregivers performed alongside a randomised controlled trial**



#### **In press as**

Sturkenboom IHWM, Nijhuis-van der Sanden MWG, Graff MJL. Process evaluation of a home-based occupational therapy intervention for Parkinson's patients and their caregivers performed alongside a randomized controlled trial. *Clinical Rehabilitation*, in press.

## Abstract

### Objective:

To evaluate fidelity, treatment enactment and the experiences of an occupational therapy intervention in Parkinson's disease (PD), to identify factors that affect intervention delivery and benefits.

**Design:** Mixed methods alongside a randomised controlled trial.

**Subjects:** These include 124 home-dwelling PD patients and their primary caregivers (recipients), and 18 occupational therapists.

**Intervention:** Ten-week home-based intervention according to the Dutch guidelines for occupational therapy in PD.

**Main measures:** Data were collected on intervention dose, protocol process, content of treatment (fidelity), offered and performed strategies (treatment enactment), and recipients' experiences. Therapists' experiences were collected through case note analyses and focus group interviews.

**Results:** Mean intervention dose was 9.3 (SD 2.3) hours. Mean protocol process adherence was high (93%; SD 9%), however the intervention did not (fully) address the goal for 268 of 617 treatment goals. Frequencies of offered and performed strategies appeared similar, apart from 'using other tools and materials' which showed a drop from 279 advised to 149 used. The recipients were satisfied overall with the intervention (mean score 8 out of 10). The therapists noted positive or negative influencing factors on both process and benefits: the research context, the socio-political healthcare context, the recipients' personal and contextual factors, and the therapists' competence.

**Conclusion:** We found some prerequisite factors in equipment provision and available dose important for treatment delivery. Other elicited factors related to, or affected, the required professional competencies and tools to tailor interventions to the complexity of interacting personal and contextual factors of patients and caregivers.

## Introduction

From 2011-2013, we conducted a clinical trial to evaluate the efficacy of home-based occupational therapy for people living with Parkinson's disease (the OTiP trial).<sup>1, 2</sup> Following the existing clinical practice guidelines,<sup>3</sup> the intervention addressed individual goals related to engagement in meaningful activities. The results showed that occupational therapy significantly improved patients' self-perceived performance in prioritised daily activities as assessed with the Canadian Occupational Performance Measure (COPM).<sup>4</sup> Although the difference in progress on the group level was significant, only one-third of the patients in the intervention group showed a clinically important improvement when using the COPM's predefined criterion for minimal clinical important change. This process evaluation focuses on the identification of relevant factors to enhancing intervention delivery and benefits in the future.

Based on the data in the efficacy study we hypothesised that not all intervention effects were captured with the COPM. This is because we found some incongruence between priorities in daily activities as assessed at baseline COPM by the researcher, and the treatment goals addressed in the intervention.<sup>1</sup> The COPM's priority specificity hinders the ability to find treatment results outside the original priorities. This means that we need insight into the perceived benefits of the OTiP intervention and into factors that contributed to or hindered the intervention delivery. Understanding these factors would help to develop implementation strategies and to improve the guidelines.

The OTiP intervention was individually tailored to suit the impact of Parkinson's disease (PD) on activities and participation, the person's priorities, and daily living context. This means that within a range of recommended strategies and approaches, the actual therapy goals and intervention strategies differed between patients (and caregivers) participating in the trial. This added to the complexity and potential variations in intervention content and delivery.

To gain insight into the various interacting components of complex interventions in a trial context, a process evaluation is essential.<sup>5-8</sup> The literature highlights several important concepts that need to be addressed in process evaluations. A central concept is 'treatment fidelity', which is defined as the extent to which the intended intervention was provided by the therapists.<sup>9,10</sup> Another concept is 'treatment enactment', the extent to which recipients (i.e. patients and caregivers) apply the interventions in daily life.<sup>9, 10</sup> Identification of the intervention's strong and weak aspects, as well as characteristics of the therapist, patients, and socio-political healthcare context that act as barriers or facilitators for the intervention may explain the level of treatment delivery or outcomes.<sup>5,7,11</sup>

With the process evaluation of the OTiP intervention we therefore aimed to elicit the following:

- a) The treatment fidelity: the dose, the protocol process adherence and content of treatment delivered compared to the protocol
- b) The level of treatment enactment by recipients
- c) The experiences of recipients with the intervention process and its outcomes
- d) The experiences of therapists on the perceived benefit of the intervention for the recipients and on the barriers and facilitators for successful treatment delivery.

This will provide insight into factors that influenced the intervention delivery and the perceived benefits in the OTiP trial, which will be used to enhance intervention delivery and benefits in the future.

## Methods

We conducted the process evaluation alongside a multicentre, randomised controlled clinical trial (the OTiP trial, 2011–2013).<sup>1,2</sup> We used a mixed methods design, combining qualitative and quantitative methods.

The trial was set within the ParkinsonNet care context in the Netherlands.<sup>12,13</sup> This is a network of specialised health professionals, who are organised in regional multidisciplinary networks.

Data were collected from all 18 occupational therapists who delivered the OTiP intervention, and from the 124 home-dwelling PD patients and 117 caregivers who entered the intervention arm of the OTiP trial. Information on the recruitment, inclusion and exclusion criteria, and attrition, as well as the baseline characteristics are reported in detail elsewhere.<sup>1</sup> Relevant data to this process evaluation include: the patients' ages, which ranged between 42 and 87 years (median 71), and a full range of disease severity, but the majority of patients (77/124; 62%) were in Hoehn and Yahr stage 1 or 2 (mild disease). Most participating caregivers (103/117; 88%) were the patient's partner.

The participating therapists were all women with a median practice experience of 12 years (range 2–28) and a median ParkinsonNet experience of 2 years (range 1–4). As members of ParkinsonNet, all OTiP therapists received at least 3 days training on treating PD patients. Prior to the OTiP trial participation, the therapists received an additional 3 days of training on clinical skills and protocol procedures. Halfway through the study, 16 of the therapists attended a 1-day booster training. To discuss issues and experiences, therapists used a secure online group platform and they individually received a median number of 5 (range 2–9) telephone coaching sessions with an expert occupational therapist (IS).

## The intervention

The OTiP intervention was a 10-week home-based, client-centred occupational therapy intervention according to the Dutch guidelines.<sup>3</sup> The maximum contact time was 16 hours.

As part of the diagnostic phase, the guidelines recommend the use of the COPM to prioritise and measure activities during which the patient experiences problems.<sup>4</sup> In the OTiP trial, the COPM was the primary outcome measure, and, hence, this measure was administered by the blinded assessors before and after the intervention. The therapists received the baseline COPM as a starting point, but they were allowed to verify and, if needed, re-prioritise issues with the patient in the diagnostic phase. Caregivers could also identify their own intervention goals related to managing the caring situation whilst considering their own well-being. Subsequently, using shared decision making principles, goals and strategies were determined for both patient and caregiver.

The intervention strategies focused on 'the person' (coaching and strategy training of recipient), 'the activity' (adaptations of activities and daily routines) and the 'environment' (e.g. assistive devices, layout, and support). Collaboration with other healthcare professionals took place as usual.

## Measures and procedures

We used a variety of sources to collect data. Table 1 presents an overview of the evaluation components and the respective sources and variables used.

Details on dose delivery (e.g. frequency and duration of sessions) were recorded by the therapists in the case notes. Data for protocol process adherence (i.e. which 12 steps of the protocol were performed and in which sequence) were extracted from a structured list in the case notes (see Appendix A). For each case, we calculated a process adherence score as the (number of steps performed)/12\*100%, and we established a sequence following the completion of diagnostic phase, treatment planning, and interventions.

Because the content of the intervention was goal-oriented and the number of goals varied between recipients, we measured the quality of the treatment content and treatment enactment for each treatment goal. Through content analysis of the case notes, the researcher (IS) determined the content indicator score by establishing agreement of recorded treatment with predefined content quality criteria based on the guidelines (see Table 3). When all criteria were met a content indicator score of 2 was given. When no criteria were met, a score of zero was given. Goals with partially met criteria received a score of 1, and the main reason for not meeting the criteria was recorded.

To assess treatment enactment, we extracted both the intervention strategies offered in treatment, and the strategies performed by the recipient as registered in

**Table 1** Overview of the evaluation components, respective data sources and process variables used

Evaluation component	Sources used	Process variables
<b>Quantitative</b>		
a) Treatment fidelity:		
Intervention dose	Case notes	Number of sessions, total direct contact time
Process and content of treatment delivery	Case notes	Protocol process adherence score Content indicator score (agreement treatment delivery with predefined content quality criteria)
b) Treatment enactment by the recipients	Case notes	Difference in strategies offered and strategies performed by recipients
c) Recipients' experiences with the intervention	Experiences questionnaire	Satisfaction with: <ul style="list-style-type: none"> <li>- overall intervention</li> <li>- number sessions and duration intervention period</li> <li>- time used for assessment</li> <li>- involving caregiver</li> </ul> Participation in: <ul style="list-style-type: none"> <li>- joint goal setting</li> <li>- tailoring of intervention</li> </ul> Satisfaction with OTiP intervention results Recommendation to others
<b>Qualitative</b>		
d) Therapists' experiences with the intervention	Focus groups Case notes process sheet	Perceived benefit for recipients Barriers and facilitators for successful treatment including positive and negative aspects of OTiP intervention.

the final evaluation with the therapist from the case notes. To allow quantification, the strategies described in the case notes were coded within predefined categories based on the guidelines.

To collect the recipients' experiences with the intervention, the patients and caregivers completed a specifically developed questionnaire with closed questions to rate experiences on satisfaction and participation in the intervention, satisfaction with the results and recommendations to others.

To gather the therapists' experiences, the therapists listed, for each recipient, barriers and facilitators to that specific intervention on a process sheet in the case notes.

After the completion of the intervention period in the study, the therapists were invited to share their experiences in one of two focus groups led by an experienced, independent focus group leader and the researcher (IS). The topic guide (see Appendix C) covered perceptions of successful treatment, barriers and facilitators for successful treatment, and experiences with the intervention protocol. The focus group discussions were audiotaped and transcribed.

### Data analysis

Using descriptive statistics (SPSS version 20), we analysed data on treatment fidelity, treatment enactment and recipients' experiences with the intervention. In addition to the analysis of protocol adherence in each case, we also recalculated and analysed protocol process adherence per protocol step at group level, using the calculation: (cases in which step was performed)/(total reported cases)\*100%. Likewise, we analysed the data on sequences at the group level. To analyse treatment enactment, we descriptively compared the differences in frequencies between strategies offered by the therapist and strategies performed by the recipients.

We analysed the data on therapists' experiences using the constant comparison method (qualitative data analysis).<sup>14, 15</sup> Texts on barriers and facilitators from the case notes and transcripts of the focus groups were coded via line by line analysis, which was supported by Atlas.ti (version 7). To establish a reliable coding structure, an initial part of the texts was coded separately by the first author (IS) and an independent research assistant. This was followed by comparison and discussion in order to reach consensus on the coding system. The coders first determined whether the text line(s) involved a result, a barrier or a facilitator and within each of these categories open coding was used. The first author (IS) then analysed the coded data to identify emerging subcategories. These were presented and subsequently grouped in main themes through discussion amongst members of the research team (IS, MG, MNvS). To check our interpretation, we sent the results of the qualitative analysis to the therapists and asked for written comments.

To enhance reliability and credibility, we compared the quantitative results of treatment fidelity and treatment enactment with the experiences of recipients and the qualitative results on the therapists' experiences to elicit convergence or dissonance between results.<sup>16</sup>

## Results

### Treatment fidelity

The results regarding intervention dose showed that participants in the intervention group received a mean treatment dose of 8.5 (SD 2.2) sessions. The mean direct intervention time per patient plus his or her caregiver was 9.3 (SD 2.3) hours.

The mean adherence indicator score per case was 93% (SD 9%). Group-level results for protocol process adherence (Table 2) revealed that therapists' adherence per protocol step was between 93% (115/123) and 100% (123/123) of cases, apart from observation of the patient's activity performance with the caregiver. This was delivered in 51% of the cases (58/114). Complete adherence (i.e. all relevant steps performed) was reached in 46% of the cases (56/123).

The intended sequence of subsequently completing diagnostic phase, treatment planning and interventions was followed in 65% of the cases (80/123).

In total, the therapists had formulated 617 goals in the treatment plans. The analysis of the content indicator score suggests that in 57% of goals (349/617), the intervention fully met the quality criteria. In 43% of the goals (264/617), the intervention did not fully meet the quality criteria (content indicator score 1). Table 3 provides the main reason for not meeting the criteria. The most frequent reason (10%; 59 of 617goals) was lack of practice in using the advised aids and adaptations in activities because these modifications were not in place. For 9% of goals (55/617), recipients' motivation for the goals or strategies changed and in another 9% (53/617) the chosen mix of strategies did not fit the problem analysis or goal.

### Treatment Enactment

The frequencies of strategies offered compared to strategies used by recipients (treatment enactment) are outlined in Table 4. For all 617 treatment goals save 'external reminders to aid cognitive processes', recipients' use of strategies was lower than what was offered by the therapists. The strategy 'using other tools/ materials' showed the largest drop from being advised in 45% of goals (279/617) to being used in 24% of goals (149/617).

### Recipients' experiences with the intervention

Overall, on a scale from 1–10, the mean grade of satisfaction with the intervention was 8.1 (SD 1.2) for patients and 7.8 (SD 1.5) for caregivers.

Table 5 provides a summary of the results of recipients' responses to ordinal questions from the experiences questionnaire. This table reveals that more than 70% of recipients perceived the frequency and period of interventions, and the level of caregiver involvement was (absolutely or merely) adequate.

**Table 2** Protocol adherence as recorded by therapists

	Performed, yes/ available*
Separate process steps	
1. Exploring meaning of activity/ roles with patient using themes of Occupational Performance History Interview-II	123/123 (100%)
2. Verifying/ clarifying patient's prioritised needs and current coping strategies	123/123 (100%)
3. Separate caregiver intake using narrative interview (using topic list in guidelines)	109/114 (96%)
4. Structured observation of patient's activity performance without caregiver involvement	117/122 (96%)
5. Structured observation of patient's activity performance with caregiver present	58/114 (51%)
6. Observation/evaluation of the physical performance context	117/123 (95%)
7. Summarize /interpret diagnostic phase using the checklist 'conclusion diagnostic phase'	121/123 (98%)
8. Collaborative goal-setting	120/123 (98%)
9. Shared decision making to determine interventions/strategies	119/123 (97%)
10. Interventions according to recommended strategies of the guidelines	118/123 (96%)
11. Evaluation of the goals with the recipients	120/123 (98%)
12. Finalizing intervention /agreement on next steps	115/123 (93%)
All steps performed (100% adherence)	56/123 (46%)
Sequences in time:	
Diagnostic phase completed before treatment planning and interventions	80/123 (65%)
Treatment planning before completing diagnostic phase	8/123 (7%)
Interventions before completing diagnostic phase	32/123 (26%)

\*Available refers to the number of case notes with a response. One of the case notes was completely missing. The analysis includes case notes of three patients who did not complete the intervention.

Almost all patients (98%; 118/120) and majority of caregivers (77%; 79/102) stated they were involved in collaborative goal setting. Most patients (90%; 106/118) were merely or absolutely satisfied with the intervention's results, although a smaller proportion (70%; 83/118) indicated their coping with daily activities had improved.

The questionnaires revealed that only a minority of caregivers (36%; 38/105) indicated there had been treatment goals directed at them as caregivers. Although a

**Table 3** Content indicator scores per goal, as assessed through content analysis of the case notes

Content quality criteria (score)	frequency (% of 617 goals)
All of content quality criteria were met (score 2):	349 (57%)
a. the mix of strategies offered fits with the goal/ problem analysis	
b. the approach of offering the intervention (e.g. instruction demonstration, practice) fits with goal/strategy	
c. sufficient try out and practice opportunities	
d. adequate multidisciplinary collaboration (relevant to the goal)	
e. timely monitoring of effect of intervention/ strategy; adjusting intervention if needed.	
Part of content quality criteria were met (score 1)	264 (43%)
Main reason for not meeting criteria:	
a. the chosen mix of strategies did not fit with the problem analysis/goal	53 (9%)
b. the approach of offering the intervention (e.g. instruction demonstration, practice) did not fit with goal/ strategy	45 (7%)
c. Insufficient try out and practice:	
i. due to lack of required equipment	59 (10%)
ii. due to change in patient or caregiver’s motivation for the goal or strategies	55 (9%)
iii. due to limited treatment period	24 (4%)
iv. due to personal reasons	16 (3%)
d. Insufficient multidisciplinary collaboration	12 (2%)
None of content quality criteria were met (score 0)	4 (1%)

Percentages do not sum up to 100% because of rounding

majority of caregivers were satisfied with the intervention’s results, 41% of caregivers (41/99) responded that their ability to cope with the caring situation had not improved or improved slightly. However, of the 38 caregivers who had their own goals, 79% (30) perceived their coping had (merely or absolutely) improved.

**Table 4** Frequencies of strategies offered compared to strategies used by the recipients

Focus	Strategy	Offered		Used	
		frequency (% of 617 goals)	frequency (% of 617 goals)	frequency (% of 617 goals)	frequency (% of 617 goals)
Person	Empowerment		95 (15%)		n/a
		Specific coaching and emotional support		32 (5%)	
		Informing others/asking for help	35 (6%)		32 (5%)
		Advice on helpful resources/support	135 (22%)		n/a
		Practice ‘normal’ motor skills	34 (6%)		27 (4%)
		Change of posture/position, use of ergonomic principles	259 (42%)		209 (34%)
Activity	Adaptation		86 (14%)		83 (13%)
		Strategy focused attention		61 (10%)	59 (10%)
		Self initiated cues (e.g. self instruction)	56 (9%)		47 (8%)
		Strategy for complex motor sequences	74 (12%)		69 (11%)
		Cognitive strategies	46 (7%)		43 (7%)
		Reducing amount of performance steps	96 (16%)		86 (14%)
Environment (physical)	Adaptation		20 (3%)		23 (4%)
		Change in day structure/timing/duration	38 (6%)		33 (5%)
		Change in nature of activity	19 (3%)		20 (3%)
		External cues to aid movement		19 (3%)	
		External reminders to aid cognitive process	279 (45%)		149 (24%)
		Using other tools/materials	119 (19%)		108 (18%)
(social)	Empowerment		96 (16%)		93 (15%)
	Performance				

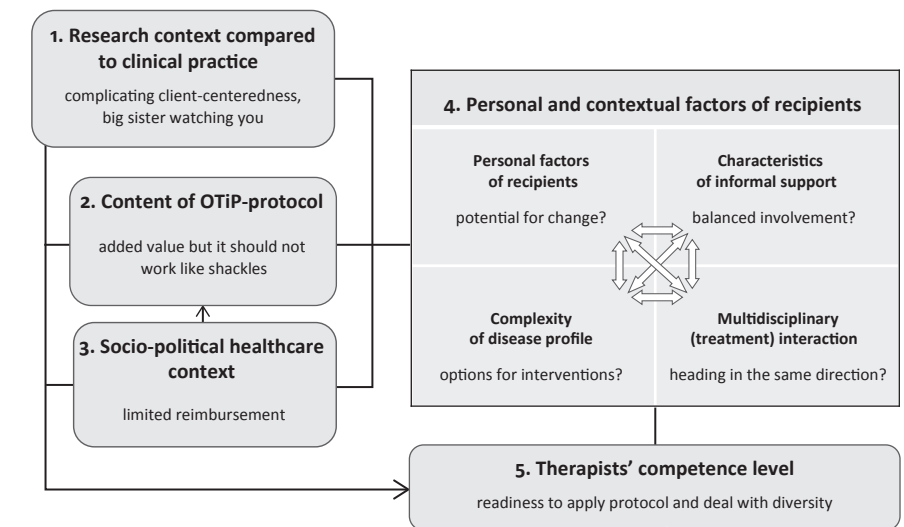
**Table 5** Recipients' experiences with the intervention

Item questionnaire	Respondents total, n	Just right	Too few/ short/ little	Too many/ long/ much
<b>Number of sessions</b>				
patients	119	94 (79%)	22 (18%)	3 (3%)
caregivers	99	75 (76%)	18 (18%)	6 (6%)
<b>Duration intervention period</b>				
patients	119	85 (71%)	23 (19%)	11 (9%)
caregivers	101	72 (71%)	20 (20%)	9 (9%)
<b>Time for assessment</b>				
patients	120	97 (81%)	5 (4%)	18 (15%)
caregivers	103	88 (85 %)	7 (7%)	8 (8%)
<b>Involvement caregiver</b>				
patients	109	96 (88%)	12 (11%)	1 (1%)
caregivers	105	89 (85%)	15 (14%)	1 (1%)
		Yes, absolutely	Yes, merely	Slightly or Not at all
<b>Participation in goal setting</b>				
patients	120	83 (69%)	35 (29%)	2 (2%)
caregivers	102	52 (51%)	27 (26%)	33 (32%)
<b>Advices fit individual situation</b>				
patients	120	69 (58%)	46 (38%)	5 (4%)
caregivers	102	52 (51%)	35 (34%)	15 (15%)
<b>Satisfaction with intervention results</b>				
patients	118	59 (50%)	47 (40%)	12 (10%)
caregivers	102	38 (37%)	41 (40%)	23 (23%)
<b>Better coping with problems</b>				
patients	118	27 (23%)	56 (47%)	35 (30%)
caregivers	99	19 (19%)	39 (39%)	41 (41%)
<b>Recommend this therapist?</b>				
patients	120	88 (73%)	19 (16%)	13 (11%)
caregivers	103	73 (71%)	17 (17%)	13 (13%)

**Therapists' experiences with the intervention**

From the perspective of the OTiP therapists, an intervention is considered successful when the recipients are satisfied with the process or results or when the therapists perceive a positive change in the recipient. The positive changes the therapists noted were in the areas of: disease insight, coping with problems, feeling about the activity (e.g. self-efficacy, pleasure), activity performance, activity pattern, communication between patient and caregiver, ability of caregiver to offer support, and space for the caregiver's own activities.

The analysis of perceptions on factors influencing intervention delivery, identified five main themes, as illustrated in Figure 1. Appendix C provides a detailed outline of the themes, underlying categories (barriers or facilitators) and supporting quotes from the focus groups or process sheets.

**Figure 1** Factors relevant in treatment delivery within the context of the OTiP trial, as elicited from therapists' experiences**Theme 1: Research context compared to clinical practice**

The therapists stated that the research procedures interfered with the intervention's client centeredness. They found it particularly difficult when the COPM priorities determined by the assessor were incongruent with the priorities that followed from their own diagnostic phase. Although a shift was allowed, the therapists were cautious



to make such changes, because they realised that the outcome of their intervention might not be measured in the research outcomes.

The therapists indicated that for a few patients, the wish to contribute to research was the main drive for participation. They felt that this drive sometimes negatively influenced the patient's motivation for setting goals and taking action.

The restricted period of 10 weeks was seen as a barrier for two reasons. First, because organizing treatment sessions of the required intensity was difficult, and second the therapists felt some strategies required a longer period to implement. Though the therapists noted that the restricted period provided clarity and focus for a few patients and caregivers.

Because of the research context, the therapists experienced a pressure (1) to deliver the intervention to the expected (high) standard within the 10-week time period, and (2) of being monitored ('big sister watching you'). Yet, they also perceived this performance pressure as a positive because having to use tools and procedures they might normally skip or avoid ensured an increase in the level of expertise when applying the guidelines. Thanks to the research, the therapists saw more Parkinson's patients in a short period, enhancing their competence.

### **Theme 2: Content of the OTiP-protocol**

The therapists perceived the protocol's steps and tools as helpful, because these provided a clear outline and facilitated working in a structured and thorough way. However, for most therapists, the separation of the assessment and treatment phases felt unnatural and sometimes inappropriate. For example when the recipients, in the perception of the therapists, expected a quick solution. Nevertheless, they unanimously recognised the value of doing a thorough diagnostic phase to fit the intervention to the person. The separate interview with the caregiver was considered beneficial even if there were no immediate goals for the caregiver. If the caregiver did not assist or supervise the patient in daily activities, then the therapists perceived the step of observing patient's performance with the caregiver as less relevant. All therapists appreciated the step of shared decision-making as a means to stimulate recipients' self-management. Yet, they noted that a few recipients did not want to take an active role in shared decision-making, instead relying on the therapist's expertise.

### **Theme 3: Socio-political healthcare context**

Although the protocol allowed an input of 16 hours over 10 weeks, the Dutch health care system reimburses a maximum of 10 hours of occupational therapy per person per year. The therapists perceived this limited reimbursement as a problem, especially if a patient had more complex needs. In the protocol, we counted on additional eligible hours of the caregivers' insurance. However, the therapists could not utilise these hours if caregivers did not have their own goals. The therapists said they were

hesitant about sending bills to recipients for their service. Additionally, the therapists noted that the restricted reimbursement and long procedures for adaptive equipment hindered implementation of the environmental compensatory strategy.

### **Theme 4: Personal and contextual factors of recipients**

This theme summarises four categories of interacting factors from the patient's context that therapists perceived as important determinants for the intervention's content, quality, and success: (1) personal factors of patient and caregiver, (2) complexity of the disease profile, (3) characteristics of informal support and (4) multi-disciplinary (treatment) interaction.

The therapists described several personal factors of the recipient that affected the scope for change. The main facilitating factors mentioned were: readiness of the patient or caregiver to identify goals; openness to advice and interventions; the person being proactive, flexible, and solution focused; and ability to change/learn. These same factors acted as a barrier in the opposite way (see Appendix C).

The therapists noted that the disease's complexity determined options for treatment. This had several aspects. First, in advanced disease stage with severe and complex symptoms, they perceived they had limited intervention options. Second, in some patients a co-morbid condition hampered application of interventions. And last, for persons with very mild PD, a few therapists indicated they struggled to select and suggest intervention options; whereas other therapists were pleasantly surprised about what they could offer these patients.

Furthermore, the extent and quality of informal support was considered an important determining factor in the treatment process. Both overprotection and too little support were viewed as barriers, whereas understanding and connectedness between patient and caregiver were perceived as facilitators. Positive involvement of caregivers in the occupational therapy process (e.g. being present, partaking in collaborative process) was helpful, especially for patients with complex needs.

Finally, the therapists indicated that involvement of other disciplines or organizations affected the success of the OTiP intervention. For various goals it was important to collaborate with other disciplines (e.g. for mobility-related goals with physiotherapy). This collaboration was sometimes hindered by a lack of availability (e.g. slow response or waiting times) or the other professional's lack of expertise. In other cases, the collaboration was good and aided goal attainment.

### **Theme 5: Therapists' competence level**

A final theme was the therapist's level of readiness to apply the protocol and deal with the diversity of all the factors that influence process and benefits. The therapists indicated a lack of confidence with the protocol early on in the study period. However, the training, supervision, and doing (experience) fuelled competence. They mentioned

specific therapeutic skills that had been important for the quality of intervention delivery. These included: adjusting communication and approach to the person, interpreting the diagnostic phase and formulating goals, providing clarity and focus, facilitating the client to be in control of the intervention process (e.g. not coming up with an immediate solution as a therapist). Some therapists struggled more with these skills than others, and some therapists indicated their level of perceived competency depended on the particular patient or caregiver.

## Discussion

This process evaluation of the OTiP intervention revealed a diverse picture of factors that affected intervention delivery and its perceived benefits.

The analysis of treatment fidelity showed that the mean dose of the treatment was lower than planned, and the protocol process adherence was high, although 43% of the treatment goals did not (fully) meet the predefined content quality criteria. Treatment enactment appeared high because the frequencies of strategies being used by recipients were similar to the ones being offered during treatment. Only the strategy 'using other tools and materials' showed a large drop in enactment because of the absence of the environmental adaptations within the treatment period. Most recipients were satisfied with the intervention and results.

The therapists expressed different factors that positively or negatively affected the intervention process and benefits. These included: the research context, the socio-political healthcare context, the recipients' personal and contextual factors and the therapists' competence.

Combining and comparing quantitative and qualitative results, the main factors relate to (1) treatment dose, (2) involvement of the caregiver, and (3) the therapist's competencies to facilitate a successful treatment.

Regarding the treatment dose, the therapists perceived the restricted period of 10 weeks mainly as a barrier. Interestingly, only a minority of recipients felt the intervention period was too short. Moreover, the mean time used (9.3 hours) indicated that not all available hours were used. Although there is very little research on optimal intensity and duration for allied health interventions for Parkinson's patients, we assumed a relatively high intensity is required for patients to acquire new skills and to keep up momentum of behavioural change. Following the feasibility study, in which the time issue also came up,<sup>17</sup> we addressed the need for prioritisation and for ensuring high intensity skills training in the therapists' training. However, the remaining bottlenecks mentioned by the OTiP therapists were the difficulty in establishing quick and efficient multidisciplinary communication and slow procedures for acquiring equipment. The latter was confirmed by the findings that 'insufficient

practice due to lack of equipment' was the most prominent reason for not reaching the highest content indicator score.

The findings suggest that the efficiency of equipment application procedures and multidisciplinary collaboration need to be enhanced. Moreover, therapists need to carefully consider the appropriate dose (intensity and total duration) depending on the strategy chosen. Nevertheless, we realise that the maximum available dose strongly depends on situational factors in countries. In the Netherlands, the reimbursement of community occupational therapy in the basic insurance is limited to 10 hours per client (patient or caregiver) per calendar year.

This process evaluation highlights the value of involving the caregiver in the intervention. The therapists perceived caregiver involvement and the quality of the patient-caregiver partnership as keys to a successful intervention, especially for more complex cases. The patients and caregivers were also positive about the extent of caregiver involvement in the intervention. The importance of including both the patient and caregiver in chronic disease management to enhance outcomes is confirmed in the literature.<sup>18</sup>

In the diagnostic phase, a separate caregiver interview allowed assessment of the caregivers' experiences and needs. This step was well adhered to, and the appreciation of 85% of the caregivers for the time taken by the therapist to assess their needs, indicates that this was a valued aspect of the total assessment. Notably, relatively few caregivers had their own goals for occupational therapy. This highlights that the role of the caregiver as facilitator for the patient to reach goals was more prominent than the role of the caregiver as a client with his or her own intervention needs. In clinical practice, therapists should be aware of the distinction in these roles and the difference in approach.

The required and perceived extent of professional competence in applying the protocol and tailoring the intervention to various relevant patient and contextual factors was another key factor elicited from the therapists' experiences. The final choice of individualised strategies and approaches was the result of complex clinical reasoning in the recipient-therapist interaction. The fidelity evaluation showed some reasons for the lower content indicator score that refer to suboptimal clinical reasoning such as inadequate selection of strategies or approaches.

Although the therapists appreciated the structure and tools in the protocol, we recognise that the OTiP intervention protocol (i.e. guidelines) only offers global guidance on clinical reasoning. Therefore, developing and implementing more specific clinical reasoning tools for different scenarios might enable therapists and recipients to improve the tailoring of interventions.

The therapists felt that 'performance pressure' and seeing a high number of patients increased their experience in using the protocol and, subsequently, their clinical reasoning. To accelerate effective clinical reasoning and the implementation

of guidelines in practice, the role of peer assessment is mentioned in the literature as a possible solution.<sup>19</sup> This might be a viable option in clinical practice. Within the organizational structure of Parkinson's care in the Netherlands, peer assessment or expertise sharing can easily be introduced because there are regular mono- and multidisciplinary meetings between specialised professionals. It might be more difficult in other countries to identify and organise peers who specialise in PD. Future research would need to review the effectiveness of these strategies in improving quality of treatment.

There are some methodological limitations to this study. First, we performed the data analyses after the trial outcomes were known and recent guidelines for process evaluation of complex interventions highlight this order's potential bias.<sup>7</sup>

Second, to minimise bias in qualitative data, we used an independent focus group leader, involved independent research assistants to establish the coding structure's validity, and verified the results of the analysis with the participating therapists. Yet, the main data analysis was conducted by members of the same research team as in the outcome evaluation. This enhanced understanding, but might have led to tunnel vision.

Third, the analysis of treatment fidelity was based on therapist-reported data. Therapists are prone to over- or underestimate aspects of their actual clinical behaviour and this impacts on the results' validity.<sup>20</sup> Literature on fidelity assessment suggests that direct observation using a rating list of observable behaviours is a more valid way of monitoring treatment fidelity and treatment enactment.<sup>9, 21</sup> Because the intervention took place in the patients' home environment it did not seem feasible to use this method. Nevertheless, use of video for assessment of therapist fidelity might be an interesting option for future trials and may also be used in peer assessments in clinical practice.

Finally, the quality of the intervention delivery was scored per treatment goal and not per case. Resultantly, we could not provide an overall content indicator score per case. Similarly, our method did not offer an opportunity to establish level of enactment per case. Establishing a robust system to measure the quality of the individualised intervention delivery and level of enactment per case will be challenging, but it would be helpful to examine a correlation of these variables with the outcome.

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### Clinical messages

Timely equipment provision, multidisciplinary collaboration, and adequate dose are important to increase treatment delivery.

Not all caregivers have their own treatment goals, highlighting the need to clearly define the caregiver's role in the intervention.

The complexity of interacting factors between personal and contextual factors of recipients put high demand on therapists' competencies to tailor interventions

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### Acknowledgements

The authors would like to thank all patients, caregivers and occupational therapists who participated in the study and shared their experiences with the intervention. We thank Professor (Em.) dr. Carla Frederiks and Wieteke van Dijk for leading a focus group discussion, and occupational therapy students Sascha Hilgen and Anne ter Haar for their assistance in organizing and transcribing the focus group discussions. Also, we thank Yvonne Veenhuizen and Jana Zajec (Radboudumc, Nijmegen) for their support in the analysis.

We acknowledge the OTiP project team members Professor dr. Bastiaan Bloem and Dr. Marten Munneke for their assistance in the design of the OTiP-trial giving the opportunity to study the complexity of occupational therapy in PD.

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## Supplementary material

### Appendix A Therapists' checklist for adherence to steps of the OTiP intervention protocol

Step	yes	no	n/a*	Session nr
<b>I Diagnostic phase (week 1-2)</b>				
1. Exploring meaning of activity/ roles with patient using themes of Occupational Performance History Interview-II				
2. Separate caregiver intake using narrative interview (using topic list in guidelines)				
3. Verifying/ clarifying prioritised needs and current coping of patient and caregiver				
4. Structured observation of patient's activity performance without involvement caregiver				
5. Structured observation of patient's activity performance with caregiver present				
6. Observation/evaluation of physical performance context				
7. Summarise/interpret diagnostic phase using checklist 'conclusion diagnostic phase'				
<b>II Treatment planning (week 2)</b>				
8. Collaborative goal-setting				
9. Shared decision making to determine interventions/strategies				
<b>III Therapeutic phase (week 3-10)</b>				
10. Interventions according to recommended strategies of the guidelines†				
11. Evaluation of goals with recipients				
12. Finalizing intervention /agreement on next steps				
n/a not applicable: e.g. no participating caregiver therefore no caregiver interview and no observation with caregiver present.				
† Summary of the Interventions in the Dutch guidelines for occupational therapy in Parkinson's: General approach: stimulating self management, coaching, informing and training skills Possible interventions directed at patients:				
- Use of alternative and compensatory strategies to improve task performance: e.g. use of cues, reorganizing complex performance sequences, focused attention, cognitive strategies like time pressure management, planning				
- Advice on optimizing daily routines and simplifying activities				
- Advice on appropriate aids and adaptations in the environment to enhance independence, efficiency and safety				
Possible interventions directed at caregivers:				
- Provision of information (impact of disease on daily functioning, possible carer support resources, aids and adaptations)				
- Training skills to support/supervise patient in daily activities				
- Coaching caregiver to consider and ensure own well being				

**Appendix B** Topic guide for exploring experiences with the intervention  
(focus group)

Topic	Probing open questions/leads	Elicitation method
Defining a successful OTiP- intervention	You have all seen several clients. For some the intervention will have been more successful than for others. But what in your perspective is a successful OTiP- intervention? - What characterised a successful intervention? - What type of results were achieved?	Brainstorm, followed by discussion
Facilitators for a successful intervention	What contributed to a successful intervention?  When spontaneous responses are limited, ask about specific categories: - Factors related to the therapist? - Factors related to the recipient? - Aspects of the intervention? - Organisational factors?	Interview, discussion
Barriers for successful intervention	What hindered an intervention to be successful?  When spontaneous responses are limited, ask about specific categories: - Therapist related factors - Recipients' related factors - Intervention content and process - Organisational factors	Interview, discussion
Experiences with the OTiP intervention protocol	What was nice and what was difficult in applying the protocol?  Comparing and contrasting both charts: - What is striking? - How did you deal with difficulties in application?	Using 'wailing wall' and 'cheering wall' (2 flip charts). Each therapist gets few minutes to individually add points to both flip charts.  After that group discussion on points noted

OTiP: Occupational Therapy in Parkinson's disease (refers to protocol)

**Appendix C** Overview of themes, categories, codes and representative quotes from case notes and focus group interviews

Theme	Category	Codes	Representative quotes
<b>1. Research context compared to clinical practice</b>			
Regarding content of intervention: 'Complicating client centeredness'	Priorities	COPM priorities do not fit season (-); COPM priorities irrelevant –change in priorities (-)	(-) 'The season really played a role in the priorities! These [priorities] had been established earlier ..and then..yes '(F1/T3). 'Then you were just in the wrong period ' (F1/T2)
	Assessment	Repetition (-)	(-) 'The predetermined priorities [by the assessor] were difficult. Goals change in time and then?' (F1/ T6)
	Period of intervention	Fixed and limited period: clarity (+); Fixed and limited period: negative (-)	(+) 'I felt the set time of 10 hours was nice for some persons. We'll work towards that and then not for a period. And then later there can be a continuation...'  (-) 'I found the 10 week period difficult, especially when you had to apply for equipment'(F2/T14) That.., and if there were 5 COPM priorities and then from the intake there were an additional 3 priorities and that had to be done in the 10 weeks. (F2/T13)'...If they would not have participated in the research you would spread the time more'(F2/T10)
Regarding involvement client 'Complicating client centeredness'	Motivation	Research participation main drive (+; -)	(-) I had a gentlemen [client] who said: "I participate for the research, but I don't experience problems" (F1/T6)
	Burden	level of burden for client (-); planning in busy diary client (-)	(-) 'Generally it is quite burdensome when all the therapist phone [for appointments]...you noticed [in OTIP study], two appointments [per week] and then they need to go to the hospital and they need to do this and that...the diaries are just full..' (F1/T2)
Regarding OT behaviour 'big sister watching you'	Performance pressure	Good to have the big stick (+); Telephone consultation with expert OT helpful (+); pressure to perform well for the research (-)	(+) '..and we had to do it now, so then...you do it..' (F1/T5)  (-) '...there was [I felt] some pressure..you could not let things go at ease. It had to happen...' (F1/T1)
<b>2. Content of the OTiP protocol</b>			
Added value but it should not work like shackles	Value of structure of OTiP process	Structure protocol helped (+); Steps are not always all necessary (-); separation assessment/treatment unnatural (-) Assessment process extensive (+/-); In own environment (+); Added value of goal evaluation (+)	(+) Because of the structure [of the protocol] you are comprehensive, you don't forget anything (F2/9). You are forced not to forget anything (F2/T12)  (+) The list of possible interventions per problem. I found that nice just to check: have I not forgotten anything...(F1/T7)  (-) Observation with caregiver: they don't do concrete activities together, only separate from each other. This had no priority. (C/T11)  (-) What I found hard was the separation [in assessment and treatment]. When you had patients who were not greatly motivated, who were thinking I don't know why we participate, you know. Then you do the observation and leave and again nothing had been done....(F2 ,T10).  (+/-) What I liked...at least, it made the success bigger to me, was the assessment phase being more extensive than I was used to, and because of that I found out more about the motivation of why they want certain things and how to draw up the treatment plan.....A drawback was that the assessment phase took a lot of time in relation to the total time available.(F2/T11)

## Appendix C Continued

Theme	Category	Codes	Representative quotes
<b>2. Content of the OTiP protocol</b>			
	Value of OTiP assessment	Added value of narrative interview (OPHI) (+); added value caregiver interview (+); Information interviews not always relevant for OT (-); Observation provided insight (+); Limited value of observation (-)	<p>(+) I noticed that with the OPHI you get better insight into what kind of person someone is. And what someone finds important. How did he usually solve problems? So also for [selecting] the direction of the solution you can then connect better with someone.. (F1/T6)</p> <p>(+) Something I did not used do that much before.., I talked to the partners separately, I talked with the caregiver separately. I notice that this provided so much information and insight, really, I found that of great value. (F2/T12)</p> <p>(+) And the observation [with caregiver present]...then you see things happening. Persons just say : I hold in, I let him do all of it. But if you watch them do it then you see: yes that is what you think, but a lot is happening in what you still do. What you are not aware of that you do...So that has added value to do an observation together. (F1/ T5)</p> <p>(-): Well.. it was just clear that I could not use what was said [in the narrative interviews], to..what I could address as an OT...but it was on the table... (F1/T1)</p>
	Value of specific OTiP (reporting) tools	Clear and helpful (+); Suboptimal (-)	<p>(+) The reporting per goal I liked very much...because you had more...overview (F1/T5))</p> <p>(+) The list of possible interventions per problem. That I found nice just to see: have I not forgotten anything...(F1/T7)</p> <p>(-) What I did not like..what I found hard...after I had used the guide [topic list for OPHI] for the interview ..and then I had to fill in the summary form of the OPHI.. and this had other headings and I had to think: what should go where?... (F1/T5)</p>
	Value of shared decision making	Added value of collaborative goal setting/ action planning (+); shared decision-making does not fit person (-); phase of goal-setting too long (-)	<p>(+) Goal setting: nice to do it this way together. The patient and caregiver also provided good suggestions for possible actions/interventions. (C/T8)</p> <p>(-) and the older clients especially think 'you are the OT' and specifically when the goals had to be SMART. (F2/T10)</p>
	Value of OTiP intervention strategies	Interventions protocol effective (+); interventions activate client (+); intervention protocol limited added value (-)	<p>(+) Cues and movement strategies had not been tried yet; there was a lot to win.(C/T3)</p> <p>(+) A lot of was an eye opener for Mrs. [the client], how you could do things differently. (C/T9)</p> <p>(-) Due to the long illness history and the course of disease, the couple had already so much experience that there was not a lot to add, sometimes just some assurance. (C/T13)</p> <p>(-) There was no solution for the problem of getting out of bed. Also when another ParkinsonNet therapist observed with me, it did not work. (C/T6)</p>
<b>3. Socio-political healthcare context</b>			
Limited reimbursement	Reimbursement	Limited reimbursement equipment (-); limited reimbursement occupational therapy (-)	<p>(-) And I noticed that sometimes we did not get the equipment reimbursed that I thought was necessary and then...eventually the client did not get it reimbursed.. I found that a limitation (F1/T6)</p> <p>(-) We cut ourselves in the fingers [not reporting extra time spend]...because you know. We keep ourselves neatly to the 10 hours because I think there are few OTs who write a bill for payment [for the client] after the 10 hours. (F1/T4)</p>

## Appendix C Continued

Theme	Category	Codes	Representative quotes
<b>4. Personal and contextual factors of recipients</b>			
Personal factors patient/caregiver: Potential for change?	Level of readiness for goal identification	Acceptance disease (+); Good insight into disease and problems (+)	(+) Mrs. [the client] knew exactly what she wanted and was flexible in the way the goal should be reached (C/T11)
		Difficulty acceptance disease/situation (-); No good insight into disease and problems (-)	(-) Mr. and Mrs. [the couple] did not want to think about the future and the consequences of Parkinson. (C/T11)
	Level of acceptance of/adherence to intervention	Able to communicate need (+); Communication of needs/goals not optimal (-); No (realistic) perception/formulation of occupational issues (-); Own (realistic) perception and formulation of occupational goals (+)	(-) [the client] Is not satisfied with the current situation, but does not see the point [of goals]. (C/T8)
		Cooperative (+); Motivated (+); Not open for advice or support (-); Not open to certain intervention/changes (-); Open for advice or support (+); Questioning added value of intervention (-); Limited trying out and practice of strategies (-); Trying out and practice of strategies(+)	(+) Mr.[the client] is clear and concrete in his goals(C/T16)
Personality traits, values and coping style	Level of acceptance of/adherence to intervention	Importance active and independent lifestyle (+); All or nothing approach (-); Positive life perspective (+); Pragmatic/acting person (+); Solution focused (+); Using humour (+); Anxiety/lack of confidence (-); Being controlling and inflexible (-); High level of perseverance (+); Ignores own needs (-); Openness in communication with OT (+); Lack of openness in communication with OT (-); Openness in communication to 'outside world' (+); No openness in communication to 'outside world'(-)	(-) Problems were mild, does not perceive problems as severe (C/T1)
			(-) The other things [strategies] he did not like [at first], he did not even want to consider these (C/T14)
	Client taking control or not		(-) [the client] had difficulty to accept aids and adaptations, especially in socially visible situations (C/T7)
			(-) He[the client] indicates to be open [for change] but he maintains his own method and asks for assistance instead of first trying himself (C/T13)
Ability to change/learn	Client taking control or not		(+) Mr. [the client] was ready to make some changes in his life and wanted some help with this. (C/T3)
			(+) Mr. wants to participate actively in life and undertakes a lot. (C/T14)
	Ability to change/learn		(-) The perception that only training hard is good. Using a gradual approach to maintain movement and endurance is not his style (C/T1)
			(-) A feeling of inferiority stops Mrs.[the client] from many opportunities (C/T11)
Ability to change/learn	Client taking control or not		(+) Mr. [the client] is very creative in thinking of solutions if you guide him in a certain direction. (C/T16)
			(-) Mr.[the client] indicates he wants to change, but he is not able to due to his personality.(C/T11)
	Ability to change/learn		(+) Coping: perseveres, wants to get more out of life. In essence she is a strong woman who only needs a little push in the back (C/T10)
			(+) Mr. and Mrs. [the couple] were very open to each other and the outside world. This enables looking forward and anticipation (C/T6)
Ability to change/learn	Client taking control or not		(-) I had expected that Mr. [the client] would give more input in setting the goals. Now I had to guide it quite a lot. (C/T8)
			(+) Both [partners] think thoroughly about limitations and possible solutions. Interventions that provide insight worked well for them. ..(C/ T14)
	Ability to change/learn		(+) He picked up instructions quickly and was able to generalise these. (C/T14)
			(-) Mrs [the client] was very impulsive, started with everything but persevering is difficult for her (C/T11)
Ability to change/learn	Client taking control or not		(-) Due to cognitive problems, some solutions did not fit [the client], he could not remember them or not use them structurally (C/T6)
	Ability to change/learn		



## Appendix C Continued

Theme	Category	Codes	Representative quotes
<b>4. Personal and contextual factors of recipients</b>			
	Contextual interference	Hindering personal situation/context (-); Facilitating personal situation/context (+)	(-) As they are going to move house in the near future he had many other issues on his mind. (C/T14)  (+) Because Mrs. [the client] had just stopped working she had more time and energy (C/T4)  (-) As she [the client] has a house full of growing up children (and is widow) she cannot always do what's best for herself, she has to compromise. (C/T16)
Complexity of disease profile Options for intervention?	Severity and complexity of PD	Mild symptoms/problems (+ / -); Severe symptoms (-)	(+) I liked that I have now seen more people with early Parkinson's.. it was nice to see they benefitted from the 'tips and tricks', with no need for equipment...(F1/T4) (-) Mr. [the client] manages activities independently. There were no 'big' interventions which we could try. (C/T8) (-) The off periods were restricting; these were unpredictable.(C/T13) (-) There are more and more bad days. (C/T16)
	Interfering co morbidity	Interfering co morbidity (-)	(-) The co morbidity of autism/compulsiveness was difficult. Due to this he dwelled in his own routines. And it was difficult to change his own convictions.(C/T4) (-) Mrs. (the client) also had knee problems and problems with her hand unrelated to PD , which made the applicability of some advices more difficult. (C/T4)
Informal support characteristics : Balanced involvement?	Extent and quality of support caregiver	Caregiver insecure/ indecisive (-); Caregiver too protective of directive (-); Due to own (health) problems unable to provide support (-); Caregiver gives time and space (+); Negative partnership/communication (-); Positive partnership/ communication (+); Caregiver provides practical support (+)	(-) The partner was fearful that Mrs. [the client] would fall and therefore rather wanted her to do 'nothing'. (C/T4)  (-) Little support/ enthusiasm from caregiver. Caregiver is cognitively not able to do so [provide support] or stimulates too much. She is unable to play a significant role. She is just incapable to do so.(C/T1)  (+) A calm, supporting, patient and realistic attitude of the caregiver (C/T14)  (+) When the partner is also willing to think along, and together, together we are seeking for solutions..then you can [discuss]...this and that is difficult for him [the client] and what can you do differently? Then the partners can work together on that. (F1/T6) (+) Mrs [the client] has a large social network that can and will help her to sustain the changes (C/T11)
	Extent and quality of social network	(Large) social network support (+); No attention for needs (-)	(-) [The client] expects attention of others. This results in disappointment because the attention is not there. (C/T1) (-) The caregiver (mother, living in) found it hard to be confronted with the disease of her daughter and was often away during the treatment sessions. Just a way not to see it I think. (C/T4)
	Involvement of caregiver in OT process.	Caregiver insufficiently involved in occupational therapy process (-); Caregiver positively involved in occupational therapy process (+)	(+) The fact they were always both present and Mrs. [the caregiver] comprehended and applied the tips.(C/T12)
Multidisciplinary (treatment) interaction Heading the same direction?	Effects of other interventions	Negative effect other interventions (-); Positive effect other interventions (+)	(-) Patient was not optimally medicated, [resulting in] one step forward, two back (C/T10)  (+) It was helpful that the patient had consultations with the psychologist in the same period (C/T3)  (+) I suggested contacting a ParkinsonNet physiotherapist. He [the client] organised this. This has also led to improvement.(C/T14)

## Appendix C Continued

Theme	Category	Codes	Representative quotes
<b>4. Personal and contextual factors of recipients</b>			
	Quality of collaboration	Good collaboration with other professionals/organizations involved (+); Suboptimal collaboration with other professionals/ organizations involved (-)	<p>(+) Quick actions for equipment that needed to be applied for and a quick response and collaboration of local government and suppliers. (C/T2)</p> <p>(+) Well the physiotherapist that I know via ParkinsonNet, that worked well and I had two [clients] with this physiotherapist. We communicated at equal level about the transfer problems. (F2/T14)</p> <p>(-) It took a lot of time to call the other disciplines involved before you had contact or would get contact at all. I found it very hindering that this was such a struggle. (F2/T10)</p> <p>(-) It was hard to start up the use of the metronome in collaboration with the respective physiotherapist.(C/T7)</p>
<b>5. Therapists' competence level</b>			
Readiness to apply protocol and deal with diversity	Level of familiarity with protocol	Feeling familiar with protocol (+); Feeling unfamiliar with protocol (-); I should have acted differently (-)	<p>'You need to know about the disease, have sufficient knowledge. (F2/T9) ' And the protocol, the more you felt familiar with this, the easier it was. Real experience with the protocol '(F2/T13)</p> <p>(-) I did not yet feel familiar with the protocol. Knowing is one thing and doing is another. I felt uncertain about myself (C/T9)</p> <p>(-) I was doing the intervention based on my own experience and fell into routines and forgot the tips in the book [guidelines/protocol] (C/T12)</p>
	Using adequate approach/strategies	Adjust communication and approach to client (+); Facilitate client to have control in process (+); Providing clarity and structure (+); Positive therapeutic relationship (+); Taking time (+); Difficulty adjusting communication and approach to client (-); Feeling uncomfortable (-); Formulating goals difficult (-); Hard to interpret assessment (-); difficult not to come with solutions immediately (-)	<p>'And the extent your conversation skills are adequate to make the person take the next steps '(F1/T7)</p> <p>(+) I gave time and opportunity [to the client] to find his own solutions although these were 'less good' from an ergonomic perspective (C/T3)</p> <p>(+) Created trust, took time, influenced the intervention process with humour, talked in their own dialect (C/T17)</p> <p>(-) I found it hard to fit the interventions of the guideline flexibly to the individual situation.(C/T9)</p> <p>(-) Because goals were not defined clear enough, it was unclear whether they had been reached or not. (C/T3)</p> <p>(-) I found it hard to determine interventions together with the client, because often I already had the interventions in my head. (C/T6)</p>

F1, F2= source focus group 1 or 2; C= case notes; T1-T18= identifier therapist; (+)= a facilitator for a successful intervention; (-) = a barrier to a successful intervention; (+/-)= indicated as both a barrier or facilitator for a successful intervention; OT= Occupational Therapist; COPM= Canadian Occupational Performance Measure; OPHI= Occupational Performance History Interview

Chapter 7

**General Discussion**



In this thesis we assessed the added value of a home-based individualised occupational therapy intervention for persons with Parkinson's disease (PD) and their primary caregivers within the multidisciplinary ParkinsonNet context. We first modelled and evaluated the feasibility of the intervention and a randomised study design before assessing efficacy, cost-effectiveness, and process (treatment delivery and experiences).

The efficacy study demonstrated that an individualised 10 week, home-based occupational therapy intervention (i.e. the OTiP intervention) was effective in improving self-perceived performance in prioritised meaningful daily activities of persons with PD at 3 months and at 6 months follow-up. A clinically important improvement was reached by one-third of participants in the intervention group. For caregivers in the intervention group, there was a small, but significant higher quality of life at 3 months than in the control group. The burden of care did not significantly differ between the intervention group and the control group.

In the economic evaluation, no significant differences in total costs were found between the groups over a 6-month time period, but there were significant and substantial savings on institutional care in the intervention group. Cost-effectiveness in favour of the intervention was only significant for the caregivers.

The process evaluation provided insight into the complexity of interacting factors affecting treatment delivery and benefits of the intervention. The results emphasised the importance of the occupational therapists' competencies to tailor intervention strategies effectively according to variations in recipients' personal and contextual factors and the role and needs of caregivers. Moreover, the factors of adequate intervention dose, timely equipment delivery and a responsive and effective multi-disciplinary team were also found to affect intervention delivery.

This was the first large-scale and comprehensive intervention study specifically on occupational therapy for persons with PD. However, as with every research study, many discussion topics and remaining knowledge gaps arise from the findings. In this general discussion, we will reflect upon the issues relevant to increasing the effectiveness, measurement, and implementation of individualised occupational therapy. Topics will include the therapists' competence, integrated care, treatment dose and adherence, caregiver interventions, and outcome measurement. We conclude with future perspectives of the OTiP intervention and specialised care, making recommendations for clinical practice and future research.

## Enhancing therapists' competence

Occupational therapists who delivered the OTiP intervention were all members of ParkinsonNet. One of the main strategies of ParkinsonNet to enhance quality of care is to build PD expertise of the professionals.<sup>1</sup> Therapists who join ParkinsonNet complete a compulsory 3-day entry-level training. The foundations for the occupational

therapists' training are the guidelines for occupational therapy in PD. Subsequently, therapists enhance their clinical expertise by experience (i.e. seeing a sufficient number of patients) and by partaking in mono- and multidisciplinary regional seminars. They can also attend national ParkinsonNet study days or congresses and share expertise and queries in online communities.

Because intervention fidelity is an important topic in an intervention study, we offered extra training and expert consultation for the occupational therapists who participated in the OTiP study. Additionally, the OTiP therapists used case notes that were specifically developed for the OTiP study and covered recording sections based on the different steps of the intervention process. Upon completion, the case notes were sent to the researcher, who could provide feedback if needed or requested. Compared to usual clinical practice in ParkinsonNet, we realise that these were extra strategies that potentially improved competency and implementation. In the focus groups, therapists noted that the extra training and telephone consultations were helpful, as was presented in chapter 6. The fact that the therapists were monitored and the outcomes were evaluated in the research context ensured 'doing the right thing at the right time' according to the protocol. Even so, it was evident from the process evaluation that despite high OTiP intervention protocol adherence, the intervention did not 'pass' the quality criteria for a large percentage of goals (chapter 6). This was partly related to difficulty in adequately tailoring interventions.

Another key finding was that not all OTiP therapists felt fully confident applying the OTiP intervention in all patients. Therapists perceived tailoring and delivering the intervention as more of a struggle for people at both ends of the disease spectrum. The therapists' confidence grew during the study because they had treated more PD patients according to the intervention protocol. The latter confirms the ParkinsonNet's principle that increasing the number of individuals seen (i.e. experience) will increase competency. As part of ParkinsonNet's quality criteria, therapists need to treat a minimum number of patients each year to stay a member of ParkinsonNet.

The issue remains that increasing the number of patients (i.e. increasing experience) may not be sufficient for enhancing competency. Other strategies may be needed. To develop these strategies, it is necessary to unravel whether the (perceived) lack of competency is attributable to a lack of knowledge, skills, attitudes, self-efficacy or facilities. We can only partly infer this from our findings. For example, our findings suggest that it was particularly difficult to generate adequate intervention options for both ends of the disease continuum. This might result from various causes. In the guidelines, recommendations are not structured according to disease stage. The reason for this is that commonly used stages of PD are based on neuro-pathological progression (stages of Braak)<sup>2</sup> or motor symptoms progression (Hoehn and Yahr stages).<sup>3</sup> However, because of individual variations in ability to adapt to the disease specific symptoms, there is no linear relation between progression of

symptoms and progression of activity imitations or participation restrictions. Accordingly, these stages were expected to be less useful as a conceptual framework to plan occupational therapy interventions. As the disease advances, there comes a point, however, that the number of possible strategies is limited. Then, intervention planning requires a great deal of ingenuity on part of the persons involved. All the more so because there is a lack of studies relevant to occupational therapy that may guide the therapists' practice for the outer ends of the disease continuum. Apart from practice-based experience, collaborative assessment and sharing of expertise with other therapists involved (e.g. physiotherapists) may inform possible solution scenarios. We noted in the process evaluation in chapter 6 that experiences with interprofessional collaboration were variable. In conclusion, the lack of facilities (i.e. no specific recommendations available in the guidelines), lack of experience and limited use of interprofessional expertise sharing may have hampered selection and tailoring of interventions in advanced stages of disease.

In the discussion of the process evaluation study, we suggested that individualised strategies might be needed to enhance therapists' competencies. Beyond just offering continued training opportunities, we believe it will be helpful for therapists to receive individual feedback on the quality of their competencies and intervention delivery. Structured peer assessment at the regional level might be a suitable option.<sup>4,5</sup> This assessment may occur between occupational therapists or between various disciplines. Another tool that can be used to enhance therapists' competency is the use of feedback from a quality monitoring system. Currently (2015/2016), implementation of an extensive national quality registration system takes place, called ParkinsonInzicht (<http://parkinsoninzicht.nl>). This registry collects data on care utilisation, care delivery (process indicators), and experiences and outcomes of care in the field of PD. Although the primary aim is to provide transparency on quality of interdisciplinary Parkinson care, it also offers opportunity for individual therapists to benchmark certain process aspects of their practice against that of all occupational therapists and consequently understand their own strengths and weaknesses.

## Towards adequate integrated care

When we evaluated the effectiveness of occupational therapy in the OTiP study, we allowed interventions of other disciplines to continue in the control and intervention groups. Thus, we looked at the value of adding occupational therapy to the usual multidisciplinary care. Considering the complex and multifaceted nature of PD, it is essential to reflect upon the relevance and quality of multidisciplinary care.

We have already highlighted the potential value of expertise sharing and collaboration in order to generate solution scenarios in 'difficult' cases. In fact, coordinated and efficient collaboration is desirable throughout the care process of

each person with PD. When the person with PD (or a caregiver) identifies problems, it needs to be decided which professionals at what echelon of care may be able to guide or assist the person in analysing and addressing said problems. If more than one professional is involved, then goals and intervention strategies ought to match. Similarly, strategies learned in therapy should ideally be reinforced in other situations (e.g. by caregiver, homecare professionals, other therapists). Essentially, care delivery should be integrated.

ParkinsonNet has used the following strategies to enhance interprofessional collaboration: (1) organizing professionals in networks of catchment areas around regional hospitals, (2) requesting each network to organise regular multidisciplinary meetings, and (3) offering each network and all ParkinsonNet professionals an online health community for discussion. Potentially, secure online health communities offer opportunities to coordinate care when there are no formal team structures.<sup>6</sup> However, we know from clinical practice that utilisation of this organizational structure in an individual case is still limited. In fact, according to the interactive map on quality of care, ParkinsonNet professionals rate patient-related interprofessional collaboration on average only 7 out of 10 when reflecting on working in ParkinsonNet. (<http://www.parkinsonatlas.nl>)

A barrier is that professional groups or individual professionals external to ParkinsonNet might be involved in the care of a person with PD. These external professionals cannot make use of the organizational facilities and multidisciplinary meetings of ParkinsonNet. This was apparent in the OTiP study, where for example the person with PD might see a physiotherapist who is not a ParkinsonNet member.

Similarly, the current structure mainly focuses on communication between professionals; the person with PD is not included. Recent e-health initiatives have attempted to tackle these shortcomings. For instance, ParkinsonNet has initiated development of digital communication networks around a person with PD in which relevant care professionals can be invited to participate and share information. This will also include the option for online videoconferencing. For people who are digitally capable and willing to take on an active role, these online strategies might offer a solution to the communication issue. However, beyond stringent data safety requirements, logistic concerns and financial constraints, there are many barriers in implementing electronic communication in health.<sup>7, 8</sup> In addition, regardless the method (in-person or online), there are challenges to interprofessional communication and integrated teamwork.<sup>9</sup> For instance, incorporating the patient as an egalitarian partner in the team and sharing data and opinions between professionals from different organizations requires a major change in working routines and professional attitudes. Joint sessions to share and learn perspectives and expertise happen occasionally, but not routinely. This connects to the issue that interprofessional collaboration seems to depend too much on the personal competency of professionals.

Some are very eager to collaborate; others are reluctant to collaborate or lack skills to do so effectively. Therefore, a practical organizational structure that enables communication is important but not sufficient. Interprofessional collaboration should be a core competency for all professionals involved in PD care. This is particularly important now that patients are encouraged to stay in their own home longer. As a result, there will be complex care situations to manage in the community and many professionals from different organizations may be involved. Fortunately, the urgency for this vital competency is shared by the professional training institutes, and there is now more emphasis on developing this competency through shared learning.<sup>10</sup>

## Increasing treatment dose and adherence

Occupational therapy interventions often involve a change in behaviour of the person with PD or the caregiver, such as a change in performance methods, routines and lifestyle. To ensure that a change is adopted and becomes part of the daily routine, a certain intervention exposure (dose) is required. However, different dose requirements may apply according to the approach taken. To improve (adapted) performance skills or to change performance methods (e.g. incorporating use of adaptive equipment or training adapted motor skills in daily activities), a specific training programme with considerable repetition of practical exercises is essential. For changes in lifestyle and to improve patient adherence to the training mentioned above, health professionals need to implement a coaching programme. For both type of programmes, it is also important that the person with PD receives regular monitoring and feedback as well as experiences progress. Within the field of occupational therapy in PD, there is no evidence thus far for the minimal required dose for interventions. We even suspect that occupational therapists generally don't systematically consider what treatment dose is optimal. For movement-related exercise and training, the recently published European guideline for physical therapy in PD provides specific dose recommendations. These indicate the need for training 3 times a week for 30 minutes for at least 3 weeks to improve skills.<sup>11</sup> For applying cognitive rehabilitation strategies, there is no information on the proper dose. In occupational therapy, performance skills training involves both motor performance strategies and cognitive strategies.

For coaching, the trans-theoretical model of behavioural change is used as a framework (i.e. motivational interviewing).<sup>12</sup> This describes behaviour change as a cyclical model of stages in which the person can be. These stages relate to developing readiness for change, action for change, and maintenance of changed behaviour. The model does not provide any guidance on time issues, such as how much time is needed for each stage or how long a person can remain in a stage. These are likely to be too individually varied.

Apart from the big gap in knowledge and evidence on a proper intervention dose, the Dutch basic health care insurance only covers home-based occupational therapy for 10 hours per person per calendar year. Within that constraint, it is obvious that the occupational therapist will not be able to supervise a lot of face-to-face training. Therefore, an acceptable and feasible home programme for practising (adapted) occupational performance is requisite. For this, there needs to be a clear and concrete agreement with the patient and caregiver on each party's responsibility within the scope of the necessary actions. Other relevant professionals may also support applying the suggested strategies in their sessions. Adherence to the action and the strategies is crucial to achieve goals. A practical model for adherence promotion strategies within occupational therapy is provided by Radomski.<sup>13</sup> In the OTiP intervention, adherence was mainly promoted by synchronizing goals and interventions with preferences, stage of change and resources/abilities of the person (through detailed assessment and shared decision-making) as well as by involving the caregiver if possible.

In case of movement-related strategies, collaboration with the physiotherapist was recommended with the aim of reinforcing strategies.

In the OTiP study, we could only extract recipients' intervention adherence from the case notes. This means we only have information on what therapists reported. From this, it appeared that treatment enactment was high for the study's participants. In the focus groups, occupational therapists mentioned that the level of uptake and adherence varied and were important mediating factors for a successful intervention. Therefore, more detailed monitoring and exploration of intervention adherence would be informative in order to refine the strategies therapists may employ.

## Reconsidering interventions for caregivers

The involvement of the caregiver in the OTiP intervention was twofold: first, as the care-giving partner and co-therapist to reach occupational performance goals of the person with PD, and second, as a client with his or her own personal needs for intervention.

It was notable that the *mean* level of care-giving burden at baseline was low among the participating caregivers in the OTiP study. We also found that only a minority of caregivers had formulated their own goals for occupational therapy. Nevertheless, most caregivers were present for (part of) the treatment sessions. This suggests that the caregiver's involvement in the OTiP intervention was mainly focused on the roles of care-giving partner and co-therapist. Although it is suggested to be true, this is an assumption because we have not formally assessed how caregivers participated in the sessions. We did, however, evaluate appreciation of the level of

involvement. The findings detailed in chapter 6 showed that, overall, caregivers and patients appreciated the level of caregiver involvement in the OTiP intervention.

The problem is the scarcity of evidence for psychosocial interventions directed at the PD caregiver.<sup>14, 15</sup> The type of caregiver interventions in the OTiP-intervention protocol had been derived from evidence-based interventions from the field of occupational therapy in dementia care.<sup>16, 17</sup> The reason was that studies suggested that caregivers of persons with dementia and PD share similar needs for assistance on care knowledge, care skills and self-care.<sup>18, 19</sup> However, we now consider the fact that the focus of needs between these diagnostic groups differs, especially in the early stages of PD. The reason for this assumption is that because of differences in their cognitive-behavioural profile, persons with dementia are likely to be more reliant on caregiver supervision and assistance in the mild stage of disease than people with PD. Consequently, caregivers of persons with dementia may require support on care skills earlier. Still, caregivers of both diagnostic groups might have information and psychosocial support needs from diagnosis onwards.

Due to current changes in health and social care, persons with PD stay in their own homes longer and this will intensify the emotional and instrumental demands on the caregivers. Therefore, we expect that the support need for caregivers will expand. In order to advance occupational therapy interventions for caregivers of persons with PD, we recommend more in-depth exploration of varying caregiver needs relevant to the domains of occupational therapy and across the PD spectrum. We expect that the severity of disease, and the caregiver's personal and contextual resources will be factors in tailoring interventions. Furthermore, there needs to be more attention paid to the complexity of the informal care network around a person. For the purpose of this research, the caregiver was identified as one person, namely the person most involved in the care of the person with PD. In most cases, this was the patient's partner. However, the reality is that emotional or instrumental support may be shared between different individuals, making the informal care network more complex. Thus, more than one caregiver may potentially be involved in the occupational therapy process. This poses challenges for efficient information sharing and collaboration between the caregivers, the person with PD and the therapist. When face-to-face contact is difficult to arrange, virtual communication can be an option. Using a personal record (paper-based or digital) might also be an alternative or an additional choice. However, as mentioned earlier in the section on integrated care, the uptake and use of electronic communication in healthcare faces many barriers.

In the OTiP intervention, addressing caregiver needs was a part of the individual intervention for the person with PD. However, this might not always be the most appropriate option. Caregivers' own personal needs might be better dealt with in a group-based intervention, as this has the added benefit of social support and modelling among caregivers. The PPEP4all (formally PEPP training) is an example of a

standardised group training that addresses competencies to reduce caregiver stress.<sup>15</sup> Additionally, discussion groups for caregivers or partners of people with PD have been set up in some regions. Occupational therapists need to be aware of those groups and suggest them as an option when relevant.

## The challenge of measuring outcomes

In the feasibility study, we used two measures in the domain of occupational performance as a primary outcome measure: a patient-reported outcome and an assessor-rated measure based on observation. The findings indicated that the patient-reported outcome (the Canadian Occupational Performance Measure; COPM) was better suited to capture the impact of the occupational therapy intervention on occupational performance, as described in chapter 2. Conceptually, the COPM ties in closely with the individualised OTiP intervention; it allows individual variation in a person's occupational performance needs and priorities, and it considers the experiences of the person with PD as the most valid outcome. For these reasons, we used only the COPM as the primary outcome measure in our main study.

We anticipated that the measured COPM priorities would be the basis of the occupational therapy goals in the intervention group. However, the results of the feasibility study indicated that priorities and occupational therapy goals were not always congruent. To increase the level of congruence, we provided extra training to assessors of the main study specifically on eliciting and verifying patients' priorities. In the training of therapists, person-centred and SMART goal setting was practised using case studies. The level of congruence in the main trial was higher than in the feasibility study, but still there were extra goals and COPM priorities for which no treatment goals were formulated and that were not taken into account in the effectiveness evaluation. As reported in chapter 6, participating therapists stated that during the diagnostic phase the patient did not always consider the COPM priorities set at baseline as the main priorities for intervention. A study on reproducibility of the COPM in patients with various diagnoses found that priorities change.<sup>20</sup> Agreement of prioritised occupational performance problems between two assessments was found to be moderate.<sup>20</sup> Apart from a real shift in priorities in the perception of the patient, we hypothesise that the personal skills, preferences, and professional perspectives of both assessors and therapists may also influence the priority setting and goal setting processes. The literature highlights the discrepancy between goals of patients and therapists.<sup>21-23</sup> Theories on goal setting underline the complexity of the process and list many factors that affect final goal formulation, such as the recognition of problems (priorities), beliefs of self efficacy, and outcome expectancies.<sup>24</sup> Additionally, the challenges in ensuring true collaborative decision-making hamper

the process of person-centred goal setting. In order to reduce the unwarranted variation in care, clear tools that better guide the process of negotiating the perspectives of the professional, the person with PD, and the caregiver may need to be developed. Subsequently, therapists need to be trained in how to adequately manage the many factors involved in goal formulation.

Nevertheless, it is likely that priorities in daily activities will always be subject to change over time. In clinical practice, the evolving priorities can be described and explained. However, when using priorities to measure outcome, this instability is undesirable. Then, it is not possible to capture all effects of the intervention. Therefore, when using a person-centred outcome measurement such as the COPM for research, it seems imperative that outcomes on new or adapted priorities or goals are also measured. A sound method for this would need to be developed. The Goal Attainment Scale (GAS) is another individualised measure used in rehabilitation,<sup>25,26</sup> but it would share a similar problem when used as an outcome measure in research. The content of the GAS would also need to be determined with the patient at baseline. Just like with the COPM, a shift in goal areas during treatment cannot be captured. An important extra biasing characteristic of the GAS is that the professional specifies the expected outcomes beforehand and this is incorporated in the GAS goal. This means that the professional's expectations influence the target to be attained.

An alternative or additional approach could be to not only focus on the content of priorities or goals, but also on the global perceived effect of the intervention in the domain of daily functioning. Currently, a general patient-reported outcome measure is being developed in the field of occupational therapy (PROM-OT). This patient-reported outcome measure aims to evaluate the role management, self-efficacy and outcome of occupational therapy.<sup>27</sup>

An additional issue was that the outcome domain of 'occupational performance' did not always capture the possible plurality of outcome domains of occupational therapy. For example, increased preparedness for future occupational performance issues was a major gain in some cases. This could not be captured by the COPM, which measures actual perceived performance in specific activities. In the OTiP study we sought to capture multidimensional outcomes by using many secondary outcome measures in the trial next to the primary outcome of occupational performance. However, this still does not do justice to the relative weights of outcome domains that may differ between individuals.

The shortcomings of a single primary outcome measure have been raised in literature on evaluation of complex interventions.<sup>28,29</sup> A single measure assumes that there is one outcome domain and a linear cause and effect. By contrast, there are often multidimensional outcomes and interconnecting factors that affect the outcome in complex interventions.<sup>30</sup> This is especially true for interventions in which the patients are active participants in their own change process. In recent years,



models have been proposed that combine several measures to result in one composite outcome score.<sup>31, 32</sup> This is an interesting concept that may also inform further development of outcome measurements in occupational therapy.

## Future perspectives

### Transitions in healthcare: a place for specialised occupational therapy care?

The Dutch healthcare system is currently implementing major changes in order to improve long term sustainability of healthcare delivery. People are urged to take more responsibility for their own health and care. They are expected to stay in their own homes longer, primarily with support of the informal network. If formal care is needed, then this should be delivered as much as possible in or near the patient's home. As a result, primary care is organised more in community teams. Another transition is the shift from disease management (one-size-fits-all) to person-centred and individually tailored care delivery. The patient should be treated as an equal partner in health.

The home-based occupational therapy intervention for people with PD fits well in these concepts. The intervention aims to enable independent living and meaningful role engagement (participation) by helping the person with PD adapt and self-manage. A person-centred approach, including attention for caregiver's needs, is integral to the OTiP intervention. The findings of the OTiP study suggest that the intervention improves occupational performance and that there were significantly lower costs related to institutionalized care in the intervention group. These are promising results and support the value of occupational therapy in light of these healthcare changes.

Another topic of discussion in healthcare is the need for specialist expertise. An advisory report of Kaljouw to the Dutch Ministry of Health on the structure of health care delivery and professional specialists for the future indicates there should be far fewer specialists grouped around diagnoses and more generalists grouped around functional profiles.<sup>33</sup> A target is to reduce the number of specialists and disciplines in healthcare. However, we consider specialist expertise to be essential for professionals in PD care. Some competencies that are critical for a successful treatment can be deemed as *general* within the discipline of occupational therapy (e.g. type of occupational therapy assessments and principles of clinical reasoning), or across disciplines (e.g. shared decision making and interprofessional collaboration). On the other hand, specific expertise in PD is essential in order to be vigilant for possible contributing symptoms and influencing contextual factors in the assessment. Moreover, expertise in PD is crucial to draft possible solution scenarios and to determine appropriate occupational therapy strategies for the patient. The OTiP

process evaluation described in chapter 6 confirmed this. The occupational therapists highlighted the value of Parkinson's expertise in tailoring and selecting intervention options. However, we assert that even within specialised networks such as ParkinsonNet, there is room for different levels of expertise. Not all therapists need to be able to deal with the most complex cases. The most important point is that professionals recognise the boundaries of their expertise and seek collaboration with relevant experts, of their own discipline or other disciplines, in a timely and coordinated fashion.

### Recommendations for clinical practice and research

The OTiP study provides initial evidence on the overall efficacy and cost-effectiveness of occupational therapy, as well as insight into the complexity of factors influencing the intervention's process and benefits. From the findings of the OTiP study and the reflections in the discussion, we make the following main recommendations:

- (1) Recommendations to improve current occupational therapy practice in PD:
  - Persons with PD should have access to occupational therapists with Parkinson's expertise if they experience problems in meaningful daily activities or participation.
  - Although further research is needed to build the evidence base, occupational therapists can still use the current guidelines for occupational therapy in Parkinson's disease as a guide to clinical practice.
  - Occupational therapists should support the person with PD and the caregiver in the adherence to intervention strategies.
  - To optimise collaboration in integrated care networks for individual patients, occupational therapists should readily share their expertise and findings with other relevant professionals involved. To this end, they should make use of all available communication systems.
- (2) Recommendations to facilitate further implementation of the OTiP intervention:
  - There should be evaluations of current practice patterns among ParkinsonNet occupational therapists and evaluations of the extent of congruence of this practice with the OTiP intervention and guidelines.
  - Identification and prioritization of the most important determinants for implementation of the OTiP intervention (guidelines) and subsequent selection of tailored implementation strategies should be required.
  - Active monitoring of the competencies of the occupational therapists and offering continued tailored training opportunities for different expertise levels should be conducted.
  - Stakeholders should be actively involved in implementation research or initiatives.

- (3) Recommendations to enhance the content and evidence base of occupational therapy in PD care:
- There should be more exploration and evidence on occupational therapy interventions in the initial stage of PD and in advanced stages of PD.
  - Further exploration of interventions that best fit the needs of caregivers (i.e. informal network) in relation to (1) ability/feasibility of supporting and assisting the person with PD and (2) addressing their own psychosocial needs is required.
  - A decision aid should be developed that can be used to select tailored strategies that fit the possibilities of the person with PD in his or her context.
  - Further research should be conducted to develop a single or composite outcome measure that can comprehensively capture the effects of individualised occupational therapy for persons with PD.
  - Persons with PD and their caregivers should be engaged not only as research participants but also as partners in the research process.

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Chapter 8

**Summary**



The aim of this thesis was to systematically assess the effectiveness a home-based occupational therapy intervention for people living with Parkinson's disease (PD) and their primary caregivers. With the results we want to contribute to the evidence base underlying occupational therapy in the field of multidisciplinary PD care.

### **Chapter 1 – General introduction**

The progressive motor and non-motor symptoms of PD, increasingly hampers daily activities and social participation.<sup>1-3</sup> The diversity and complexity of needs of a person with PD and his or her caregivers warrant a patient-centred and multidisciplinary care approach.<sup>4-7</sup> Within this approach, medical management is complemented with input from allied health professionals who focus on improving daily functioning. In the Netherlands, multidisciplinary PD care is organized in ParkinsonNet.<sup>8, 9</sup> This is a nationwide network that comprises 66 regional networks of dedicated and trained health professionals in the field of PD.

The additional role of occupational therapy in PD care is to enable patients to engage in meaningful roles and tasks/activities in the home and community.<sup>5, 10, 11</sup> However, scientific evidence to support or refute the contribution of occupational therapists is lacking.<sup>11, 12</sup>

An important obstacle in setting up clinical trials was a lack of best-practice guidelines for occupational therapy in PD. In 2008, we developed national Dutch guidelines for occupational therapy in PD to improve uniformity and quality of care.<sup>13</sup> The International Classification of Functioning model<sup>14</sup> and the Person-Environment-Occupation Performance model<sup>15</sup> underpin the theoretical framework of the guidelines. The guidelines provide graded practice recommendations on (1) referral indications, (2) person-centred and occupation based assessment methods, and (3) various strategies (mainly compensatory) to enable activity performance and participation.<sup>13</sup> The focus is on addressing the needs of persons with PD, as well as the needs of their primary caregivers.

We do not know, however, what the effectiveness is of occupational therapy according to these guidelines within the context of multidisciplinary care. This is what we aimed to assess in this thesis.

The chapter concludes with an outline of this thesis. In our studies we first modelled and evaluated the feasibility of the intervention and a randomised study design before assessing efficacy, cost-effectiveness, and process (i.e. treatment delivery and experiences).

## Chapter 2 – Feasibility of the Occupational Therapy in Parkinson’s disease study

The second chapter in this thesis describes the initial small-scale study aimed to evaluate the feasibility of intervention delivery, the procedures of a randomised controlled trial and evaluation of the potential effect of occupational therapy for people with PD and their caregivers.

We conducted an exploratory randomised controlled trial with allocation of intervention 2:1 control, and an assessor-blinded measurement at three months followed by a qualitative evaluation of the intervention procedures and outcome. The qualitative evaluation had a phenomenological design exploring perceptions of the intervention procedures and benefits using individual interactive interviews with participants (patients, caregivers, therapists), and focus group discussion with therapists. From October 2009 to February 2010 we recruited participants from four neurology outpatient departments in different regions in the Netherlands. Eligible patients had idiopathic PD, lived at home, and reported difficulties in daily activities (covering self-care, domestic activities, work or leisure). Their primary caregivers were asked to participate if they could provide assistance for at least twice a week. The intervention involved ten weeks of home-based occupational therapy according to the Dutch guidelines of occupational therapy in PD versus no occupational therapy in the control group.

The process evaluation measured accrual, drop-out, intervention delivery and intervention protocol adherence. Primary outcome measures of patients assessed daily functioning using the Canadian Occupational Performance Measure (COPM) and the Assessment of Motor and Process Skills. Primary outcome for caregivers was caregiver burden using the Zarit Burden Inventory. Participants’ perspectives of the intervention were explored using questionnaires and in-depth interviews.

Forty-three patient–caregiver dyads were included ( $n = 29$  patient–caregiver dyads in the intervention group;  $n = 14$  in the control group). The inclusion rate was 23% (43/189) and drop-out of 7% (3/43). Full intervention protocol adherence was 74% (20/27), but only 60% (71/119) of baseline COPM priorities were addressed in the intervention. The outcome measures revealed negligible to small effects in favour of the intervention group. Almost all patients and caregivers of the intervention group were satisfied with the results of the intervention. They perceived: ‘more grip on the situation’ and used ‘practical advices that make life easier’. Therapists were satisfied, but wished for a longer intervention period.

We concluded that the positive perceived impact of occupational therapy warrants a large-scale trial. The results of the feasibility study could be used to inform the design of a large scale definite trial. We suggested that adaptations in instructions and training were needed to use the COPM as primary outcome measure.

## Chapter 3 – Protocol for the Occupational Therapy in Parkinson’s disease study (OTiP study)

Chapter 3 describes the study protocol of an adequately powered trial to evaluate the effectiveness, cost-effectiveness and process of the home-based occupational therapy intervention.

The design was a multicenter, assessor-blinded, two-arm randomised controlled clinical trial, with evaluations at 3 and 6 months. To achieve adequate power, we planned to recruit 192 home-dwelling persons with PD, who reported difficulties in daily activities. If available, the primary caregiver was asked to participate as well. The patients (patient-caregiver dyads) were randomly assigned (2:1) to the intervention or control group by a computer generated minimisation algorithm.

Patients and their caregivers in the experimental group received ten weeks of home-based occupational therapy according to recent Dutch guidelines. The intervention was delivered by occupational therapists who had been specifically trained to treat patients according to these guidelines. Participants in the control group did not receive occupational therapy during the study period.

The primary outcome for the patient was self-perceived performance in daily activities at 3 months, assessed with the COPM-performance score (score: 1–10). Secondary endpoints included: objective performance of daily activities, self-perceived satisfaction with performance in daily activities, participation, impact of fatigue, proactive coping skills, health-related quality of life, overall quality of life, health-related costs, and effectiveness at six months. All outcomes at the caregiver level were secondary, including self-perceived burden of care, objective burden of care, proactive coping skills, overall quality of life, and care-related costs. Effectiveness was assessed using a covariance analysis of the difference in outcome at three months. Alongside the efficacy trial, an economic evaluation from a societal perspective was planned, as well as a process evaluation.

## Chapter 4 – OTiP efficacy study

Chapter 4 reports on the efficacy results of the OTiP study. The study was conducted according to the planned protocol described in chapter 3, apart from a change in the method for the analysis of between-group differences. Data on efficacy were analysed using linear mixed models for repeated measures and the intention-to-treat principle. Between April 2011, and Nov 2012, 191 patients were randomly assigned to the intervention group ( $n=124$ ) or the control group ( $n=67$ ). In the intervention group 117 (94%) of 124 patients and in the control group 63 (94%) of 67 had a participating caregiver. At baseline, the median score on the COPM-P was 4.3 (IQR 3.5–5.0) in the intervention group and 4.4 (IQR 3.8–5.0) in the control group. At 3 months, these scores were 5.8 (IQR 5.0–6.4) and 4.6 (IQR 3.8–5.5), respectively. The baseline adjusted mean difference compared to controls was 1.2 (95%CI 0.8 to 1.6;  $p<0.0001$ )

at 3 months (primary endpoint) and 0.9 (95%CI 0.5 to 1.3;  $P < 0.0001$ ) at 6 months. A clinically important improvement was reached by one-third of the intervention group. The outcome 'performance satisfaction' showed similar significant effects. The intervention had no to minimal effect on other secondary patient's outcomes and caregiver's outcomes. There were no adverse events associated with the study.

We concluded that the home-based, individualised occupational therapy intervention led to an improvement in self-perceived performance in daily activities in persons with PD. Further studies were recommended to identify which factors related to the patient, environmental context, or therapist might predict which patients are most likely to benefit from occupational therapy.

### Chapter 5 – OTiP economic evaluation

Chapter 5 presents the economic evaluation over a 6-month period for both arms of the OTiP study.

This study had the same participants as described in the efficacy study detailed in chapter 4. Costs were assessed from a societal perspective including healthcare use, absence from work, informal care, and intervention costs. Health utilities were evaluated using EuroQol-5d. We estimated cost differences and cost-utility using linear mixed models and presented the net monetary benefit at different values for willingness to pay per quality-adjusted life-year gained.

In our primary analysis, we excluded informal care hours because of substantial missing data for this item. The estimated mean total costs for the intervention group compared to controls were €125 lower for patients, €29 lower for caregivers, and €122 higher for patient-caregiver pairs (differences not significant). The mean cost of the OTiP intervention itself was 760 euro. Results for cost differences in separate cost categories suggested that this was mainly compensated by a significant and substantial saving on institutional care in the intervention group (€1,458;  $P = 0.04$ )

At a value of €40000 per quality-adjusted life-year gained (reported threshold for PD), the net monetary benefit of the intervention per patient was €305 ( $P = 0.74$ ), per caregiver €866 ( $P = 0.01$ ) and per patient-caregiver pair €845 ( $P = 0.24$ ).

Occupational therapy did not significantly impact on total costs compared to usual care. Positive cost-effectiveness of the intervention was only significant for caregivers.

The study highlighted the challenge of efficient and reliable measurement of costs when conducting economic evaluations from a societal perspective.

### Chapter 6 – OTiP process evaluation

Chapter 6 presents methods and results of a detailed process evaluation of the occupational therapy intervention as delivered in the OTiP study.

In the OTiP process evaluation we aimed to elicit (1) the treatment fidelity: the dose, the protocol process adherence, and content of treatment delivered compared to the protocol; (2) the level treatment enactment by recipients; (3) the recipients' experiences with the intervention process and its outcomes; (4) the therapists' experiences on the perceived benefit of the intervention for the recipients and on the barriers and facilitators for successful treatment delivery.

We used a mixed methods design, combining qualitative and quantitative methods.

We collected data from all 18 occupational therapists who delivered the OTiP intervention, and from the 124 home-dwelling patients with PD and 117 caregivers who entered the trial's intervention arm. The data for the process evaluation included: (1) dose, protocol process, and content of treatment taken from case notes; (2) offered and performed strategies taken from the case notes; (3) recipients' experiences gathered through questionnaires; (4) experiences of therapists gathered through case note analysis and focus group interviews.

The mean intervention dose was 9.3 (SD 2.3) hours. Mean protocol process adherence was high (93%; SD 9%), while for 268 of 617 treatment goals the intervention did not (fully) address the goal. Frequencies of offered and performed strategies appeared similar, apart from 'using other tools and materials' which showed a drop from 279 advised to 149 used. The recipients were overall satisfied with the intervention (mean scores 8 out of 10). Only 1/3 of caregivers had own treatment goals. The OTiP-therapists were generally positive about the intervention protocol. Though, they noted positive or negative influencing factors on both process and benefits: the research context, the socio-political healthcare context, the recipients' personal and contextual factors, and the therapists' competence. In sum, we found some prerequisite factors in equipment provision and available dose important for treatment delivery. Other elicited factors relate to, or impact on, the required professional competencies and tools to tailor interventions to the complexity of interacting personal and contextual factors of patients and caregivers.

Based on the findings we suggest that timely equipment provision, multidisciplinary collaboration, and adequate dose are important to increase treatment delivery. In the intervention, the roles of caregiver as facilitator of the patient, or as client with his or her own intervention needs, need to be distinguished. The results also highlight the importance of adequate competencies of occupational therapists to tailor intervention strategies effectively according to variation in patient and contextual factors.

## Chapter 7 – General discussion

Chapter 7 provides a reflection on the overall findings of the studies and subsequent suggestions for clinical practice and for future research in the field of occupational therapy in PD.

The OTiP study was the first robust trial evaluating the value of occupational therapy for people living with PD and their caregivers. Although significant effects were found for perceived occupational performance, we assert there is need and scope for improving the quality and effectiveness of the intervention.

A key issue we found was that therapists sometimes struggled to select and tailor the interventions to adequately address the treatment goals. They also expressed lack of confidence in applying the protocol especially just after the start of the research. In clinical practice, we think competencies may be enhanced when therapists see sufficient patients, participate in continued training and receive individual feedback on the quality of their competencies and intervention delivery by using a form of peer assessment.

A second issue was the difficulty in realizing efficient interprofessional communication and integrated care. Although ParkinsonNet offers several organizational facilities to enhance communication between professionals, these do not always incorporate all players involved. Furthermore, interprofessional collaboration around a patient seems to depend too much on the personal competency of professionals. We suggested that enhancing integrated care and interprofessional collaboration are important areas to address.

A third issue we discussed is the need for adequate intervention exposure and intervention adherence of the person with PD. Occupational therapy interventions often involve a change in behaviour of the person with PD or the caregiver. To ensure a change is adopted and becomes part of a daily routine, a certain intervention exposure (dose) is needed. Though, different dose requirements may apply according to the approach taken: training to change performance methods or coaching to change routines and life style. With regards to determining an adequate dose, there is no evidence available thus far within the field of occupational therapy in PD. This would need further exploration. We gained limited insight into the extent that interventions were adhered to by the recipients. From the OTiP case notes it appeared treatment enactment was high. However, in the focus groups occupational therapists mentioned that the level of uptake and adherence varied and they perceived adherence as an important mediating factor. More detailed monitoring and exploration of intervention adherence, would be informative for refining and implementing adherence promoting strategies.

Next, the need to reconsider the caregiver interventions - as currently described in the guidelines - was discussed. Although therapists considered the caregiver's involvement to be facilitating for the intervention of the person with PD, only a

minority of caregivers had formulated their own goals for occupational therapy. It seems warranted to explore in more depth the varying needs of caregivers relevant to the domains of occupational therapy and across the PD spectrum. In addition, it should be considered that addressing caregiver's needs as part of the individual intervention of the person with PD might not always be the most appropriate option.

We also reflected on the challenge of measuring the outcome of an individualised intervention such as the OTiP intervention. In the OTiP study, we used the COPM, which is an individualised measure based on self-identified priorities in daily activities. Since priorities of the person with PD may change over time, the effects of the intervention on changed priorities is not measured. This highlights the need for further research to develop a single or composite outcome measure that would be more stable in capturing the effects of individualised occupational therapy for persons with PD.

Finally, I put our results in the perspective of current transitions in healthcare. Nowadays, people are urged to take more responsibility for their own health and refrain from formal care as long as possible. Another transition is the shift from disease management (one-size-fits-all) to person-centred and tailored care delivery. The home-based occupational therapy intervention for people with PD fits well in these concepts. In the discussion of reducing specialisation in healthcare, I take an opposite stance. Our findings suggested the need for specialist knowledge, skills and expertise of occupational therapists and other disciplines in PD, in order to tailor appropriate interventions.

Based on our results we summarised some future directions in three areas:

- (1) Recommendations to improve current occupational therapy practice in PD:
  - Persons with PD should have access to occupational therapists with Parkinson's expertise if they experience problems in meaningful daily activities or participation.
  - Although further research is needed to build the evidence base, occupational therapists can still use the current guidelines for occupational therapy in Parkinson's disease as a guide to clinical practice.
  - Occupational therapists should support the person with PD and the caregiver in the adherence to intervention strategies.
  - To optimise collaboration in integrated care networks for individual patients, occupational therapists should readily share their expertise and findings with other relevant professionals involved. To this end, they should make use of all available communication systems.
- (2) Recommendations to facilitate further implementation of the OTiP intervention:
  - There should be evaluations of current practice patterns among ParkinsonNet occupational therapists and evaluations of the extent of congruence of this practice with the OTiP intervention and guidelines.



- Identification and prioritization of the most important determinants for implementation of the OTiP intervention (guidelines) and subsequent selection of tailored implementation strategies should be required.
- Active monitoring of the competencies of the occupational therapists and offering continued tailored training opportunities for different expertise levels should be conducted.
- Stakeholders should be actively involved in implementation research or initiatives.
- (3) Recommendations to enhance the content and evidence base of occupational therapy in PD care:
  - There should be more exploration and evidence on occupational therapy interventions in the initial stage of PD and in advanced stages of PD.
  - Further exploration of interventions that best fit the needs of caregivers (i.e. informal network) in relation to (1) ability/feasibility of supporting and assisting the person with PD and (2) addressing their own psychosocial needs is required.
  - A decision aid should be developed that can be used to select tailored strategies that fit the possibilities of the person with PD in his or her context.
  - Further research should be conducted to develop a single or composite outcome measure that can comprehensively capture the effects of individualised occupational therapy for persons with PD.
  - Persons with PD and their caregivers should be engaged not only as research participants but also as partners in the research process.

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Chapter 9

**Nederlandse samenvatting**



Dit proefschrift beschrijft en bediscussieert de wetenschappelijke evaluatie van de effecten van een ergotherapiebehandeling aan huis voor mensen met de ziekte van Parkinson en hun directe naasten.

In dit hoofdstuk geef ik een lekensamenvatting van het gehele proefschrift. Voor een meer specifieke samenvatting van methoden en resultaten verwijs ik naar de Engelstalige samenvatting van hoofdstuk 8.

### **Hoofdstuk 1- Algemene introductie**

De ziekte van Parkinson is een voortschrijdende hersenaandoening, waarbij patiënten in toenemende mate last krijgen van diverse symptomen die het bewegen, het denken en vele andere lichaamsfuncties negatief beïnvloeden. Deze symptomen bemoeilijken het uitvoeren van dagelijkse activiteiten en sociale participatie. De complexiteit en tegelijk individuele variatie van de gevolgen van de ziekte vragen om een geïndividualiseerde en multidisciplinaire aanpak.

Er is nog geen genezing mogelijk voor de ziekte. Daarom richt de medische zorg zich vooral op het onderdrukken van de ziekteverschijnselen en het verminderen van complicaties. Paramedische zorgverleners zoals fysiotherapeuten, logopedisten en ergotherapeuten richten zich op het begeleiden van parkinsonpatiënten en hun naasten in het verminderen van de gevolgen van de ziekte op het dagelijks leven. In Nederland is de multidisciplinaire zorg georganiseerd in ParkinsonNet. Dit is een landelijk netwerk van zorgverleners die gespecialiseerd zijn in het behandelen en begeleiden van parkinsonpatiënten.

Binnen de parkinsonzorg richten ergotherapeuten zich specifiek op het benutten en vergroten van de mogelijkheden van patiënten om betekenisvolle activiteiten uit te (blijven) voeren in hun eigen omgeving. Door gebrek aan studies was er echter geen bewijs voor de toegevoegde waarde van ergotherapie.

Een belangrijke hindernis in het opzetten van studies was het gebrek aan richtlijnen voor de inhoud van de ergotherapeutische zorg bij de ziekte van Parkinson. Om de eenduidigheid en kwaliteit van zorg te verbeteren hadden we ons daarom eerst gericht op het systematisch ontwikkelen van een landelijke praktijkrichtlijn. Deze richtlijn was gereed in 2008. De richtlijn geeft aanbevelingen voor een persoonsgerichte ergotherapiebehandeling vanuit een biopsychosociaal en handelingsgericht denkkader. Binnen deze ergotherapiebehandeling wordt samen met de persoon met de ziekte van Parkinson en diens naasten, naar passende oplossingen gezocht voor ervaren problemen in dagelijkse activiteiten. Afhankelijk van de persoonlijke situatie kan een patiënt vaardigheden trainen, specifieke compensatiestrategieën leren gebruiken of kunnen aanpassingen gerealiseerd worden in de activiteit of omgeving.

We wisten echter niet of een ergotherapiebehandeling volgens deze richtlijn daadwerkelijk het dagelijks functioneren van mensen met die ziekte van Parkinson verbetert. Dit was de aanleiding voor de onderzoeken in dit proefschrift.

In onze onderzoeken hebben we eerst de haalbaarheid van de interventie en de studieopzet onderzocht. Vervolgens hebben we achtereenvolgens de effectiviteit, de kosteneffectiviteit en het proces (uitvoering van de interventie en ervaringen) geëvalueerd.

### Hoofdstuk 2- Haalbaarheidsstudie

De haalbaarheidsstudie was een kleine studie om te evalueren of ons vooropgestelde plan voor de interventie en studieopzet wel uitvoerbaar was, en om ervaringen en uitkomsten te exploreren. We hadden 43 deelnemers, die op basis van loting werden verdeeld over een interventiegroep en een controlegroep. Om mensen te werven werden mensen met de diagnose 'ziekte van Parkinson' vanuit poliklinieken neurologie in 4 regio's van ParkinsonNet aangeschreven met informatie over de studie en een open uitnodiging tot deelname. Deelnemers woonden thuis en ervoeren problemen in dagelijkse activiteiten. Hun meest betrokken naaste werd ook gevraagd deel te nemen aan de studie.

De mensen in de interventiegroep ontvingen 10 weken ergotherapie aan huis volgens de richtlijn; mensen in de controlegroep ontvingen geen ergotherapie.

We inventariseerden de ervaringen met de interventie en het onderzoek aan de hand van: (1) individuele interviews met de zeven betrokken ergotherapeuten en van iedere therapeut één deelnemende patiënt en mantelzorg, (2) een focusgroep interview met de ergotherapeuten, en (3) vragenlijsten bij alle deelnemers in de interventiegroep.

Met verschillende meetinstrumenten brachten we aan het begin en na 3 maanden het dagelijks functioneren van de patiënt en de ervaren zorglast van mantelzorgers in kaart. De uitkomsten van deze metingen werden vergeleken tussen beide groepen.

De resultaten gaven aan dat de onderzoeksprocedures grotendeels haalbaar waren, hoewel we relatief veel mensen moesten aanschrijven om voldoende deelnemers te krijgen (23% van aangeschrevenen deden mee, 7% vielen uit tijdens de studie). Bij 74% van de deelnemers volgden de therapeuten alle stappen van het behandelprotocol. Bijna alle deelnemers waren tevreden over de behandeling en de behaalde resultaten. Velen benoemden dat het uitvoeren van activiteiten of het bieden van zorg was verbeterd door het toepassen van praktische adviezen en nieuwe strategieën. Tevens gaven ze aan meer grip te ervaren op hun situatie door een vergroot inzicht in wat de ziekte betekent voor het dagelijks functioneren. De therapeuten waren tevreden over de inhoud van het behandelprotocol, maar vonden de behandelperiode vaak te kort. De resultaten op de uitkomsten van de meetinstrumenten suggereerden een positieve trend ten gunste van de groep die ergotherapie had gehad, maar dit kon statistisch gezien nog teveel op toeval berusten.

We concludeerden dat een grootschalig onderzoek gewenst was om de hypothese van effectiviteit van de interventie ook echt te kunnen toetsen. De resultaten van de haalbaarheidsstudie konden gebruikt worden om de keuze van meetinstrumenten nader te bepalen en de training van de therapeuten en procedures van de vervolgstudie aan te scherpen.

### Hoofdstuk 3 – Protocol voor “de OTiP studie”

Dit hoofdstuk beschrijft het geplande ontwerp van de vervolgstudie, “de OTiP studie” die de effectiviteit en kosteneffectiviteit van de ergotherapiebehandeling moest kunnen toetsen.

De opzet was een gecontroleerde studie waarin na drie en na zes maanden de uitkomsten van deelnemers in een interventiegroep en controlegroep met elkaar vergeleken werden. De metingen werden uitgevoerd door onderzoeksassistenten die onbekend waren met de groepstoekenning van de deelnemers. De studie vond plaats in verschillende regio's van ParkinsonNet in Nederland en beoogde 192 thuiswonende parkinsonpatiënten te werven en, indien beschikbaar en bereid, met mantelzorg.

Deelnemers in de interventiegroep ontvingen, gedurende de eerste 3 maanden, 10 weken ergotherapie aan huis (maximaal 16 uur) gebaseerd op de aanbevelingen uit de richtlijn Ergotherapie bij de ziekte van Parkinson. Dit betekende dat de ergotherapeut na een grondige analyse van de prioriteiten, beperkingen en mogelijkheden, de patiënt begeleidde in het toepassen van strategieën of het realiseren van aanpassingen in de omgeving met het doel activiteiten beter uit te kunnen voeren. Waar dit relevant was, werd de mantelzorg betrokken bij de behandeling van de patiënt en was er ook aandacht voor de hulpvraag van de mantelzorg zelf. In de periode tussen de vervolgmetingen van 3 en 6 maanden ontvingen deze deelnemers geen ergotherapie. De deelnemers in de controlegroep kregen gedurende de gehele studieperiode geen ergotherapie.

De variabele, waar het effect primair op getoetst werd, was ervaren uitvoering van dagelijkse activiteiten na 3 maanden, gemeten met de COPM-uitvoeringsscore (score: 1–10). Andere variabelen die gemeten werden betroffen: uitvoering van activiteiten, ervaren tevredenheid met de uitvoering van activiteiten, participatie, invloed van vermoeidheid, copingstrategieën, kwaliteit van leven, aan de ziekte gerelateerde kosten, en effectiviteit na 6 maanden. Alle uitkomstmaten gericht op de mantelzorg waren van secundair belang voor de studie en omvatten de domeinen: ervaren zorglast, uren zorg, copingstrategieën, kwaliteit van leven en mantelzorg gerelateerde kosten. Om de effectiviteit te evalueren werden de uitkomsten van de beide groepen met elkaar vergeleken. Met de gegevens over effectiviteit en zorgkosten werd de kosteneffectiviteit geanalyseerd. Tevens werd er een proces evaluatie gepland voor de interventiegroep.

#### Hoofdstuk 4 – Resultaten van de effectiviteitsstudie

Het onderzoek was grotendeels uitgevoerd conform het voorafgestelde plan (hoofdstuk 3). In totaal zijn 191 patiënten willekeurig verdeeld over de interventiegroep (124 deelnemers) en de controle groep (67 deelnemers).

De leeftijd van de patiënten varieerde van 42 tot 87 jaar en de ziekte-ernst varieerde van mild tot ernstig. Er deden 180 mantelzorgers mee. De deelnemende patiënten die ergotherapie hadden gehad (interventiegroep) hadden gemiddeld een betere ervaren uitvoering en tevredenheid in dagelijkse activiteiten na drie maanden dan mensen die geen ergotherapie hadden gehad. Dit verschil ten gunste van de interventiegroep was, in iets kleinere mate, nog steeds aanwezig bij de vervolgmeting van 6 maanden. De mantelzorgers van de interventiegroep rapporteerden direct na de behandelperiode een betere ervaren kwaliteit van leven. Op andere uitkomstmaten van patiënten en mantelzorgers waren de verschillen tussen de groepen niet duidelijk genoeg.

We concludeerden dat een geïndividualiseerde ergotherapiebehandeling aan huis de ervaren uitvoering van activiteiten van patiënten verbetert, evenals de tevredenheid daarmee. Omdat er veel individuele variatie was in de mate van effect, adviseerden we dat meer studies nodig zijn om uit te zoeken welke factoren bijdragen aan een succesvolle behandeling.

#### Hoofdstuk 5- Economische evaluatie van de interventie

Voor de kostenanalyse zijn alle kosten vanuit een maatschappelijk perspectief meegenomen. Deze kostenposten betroffen: zorggebruik (consulten, opnames, hulpmiddelen), werkverzuim, mantelzorg en de kosten voor de ergotherapiebehandeling. De kosten werden geanalyseerd over een periode van 6 maanden.

Voor de kostenpost 'mantelzorger' waren er zoveel missende gegevens dat deze uiteindelijk niet is meegenomen in de voornaamste analyses. De totale kosten van de experimentele en controlegroep verschilden niet duidelijk van elkaar. Voor een aantal losse kostenposten was er wel een duidelijk verschil in kosten. Zo had alleen de interventiegroep de kosten voor de ergotherapiebehandeling en die lagen gemiddeld op 760 euro per patiënt. Daarentegen was er in de interventiegroep per patiënt een gemiddelde besparing van 1458 euro op institutionele zorg.

Om de kosteneffectiviteit (oftewel doelmatigheid) van de behandeling te bepalen werden de kosten in verhouding gezet met de uitkomsten op een standaardmaat voor kwaliteit van leven. Voor de ziekte van Parkinson geldt een afkappunt van 40.000 euro kosten per op kwaliteit gecorrigeerd levensjaar om te spreken van een doelmatige interventie. In de OTiP studie bleek de ergotherapiebehandeling op dat afkappunt doelmatig te zijn voor de mantelzorger. Voor de patiënt en de patiënt-mantelzorger koppels waren de gemiddelde resultaten wel positief ten gunste van

doelmatigheid van de behandeling maar dit was statistisch niet significant (d.w.z. kon nog in grote mate op toeval berusten).

De conclusie van de studie was dat de behandeling kostenneutraal gegeven kan worden: het bespaart in totaal geen geld, maar het kost ook niet meer. De doelmatigheid van de behandeling is niet eenduidig aangetoond.

#### Hoofdstuk 6 - Procesevaluatie van de interventie

Het doel van de procesevaluatie was inzicht te krijgen in factoren die direct of indirect invloed hebben op de kwaliteit van de behandeling en de ervaren uitkomsten.

We hebben met verschillende methoden gegevens verzameld bij alle deelnemers van de interventiegroep en bij de 18 ergotherapeuten die de behandelingen voor het onderzoek uitgevoerd hadden. De verzamelde gegevens moesten een beeld geven van: de mate waarin de behandeling was uitgevoerd volgens het protocol, de mate waarin de adviezen en strategieën die aan de patiënten en mantelzorgers geboden waren ook gebruikt werden (opvolging), de ervaringen van de deelnemers, en de ervaringen van de therapeuten.

De resultaten gaven aan dat de therapeuten minder dan de maximaal beschikbare behandeltime hadden gebruikt. Gemiddeld was er een goede opvolging van de stappen van het protocol door de therapeuten. Toch was er aan 268 van de in totaal 617 behandeldoelen niet volgens alle gedefinieerde kwaliteitscriteria gewerkt. De opvolging van strategieën en adviezen van deelnemers leek goed, behalve voor het gebruik van hulpmiddelen. De deelnemers waren over het algemeen heel tevreden over de interventie en de behaalde resultaten. De therapeuten gaven veel factoren aan die de behandeling en ervaren uitkomsten in positieve of negatieve zin beïnvloedden. Deze konden worden samengevat in vijf domeinen: (1) de onderzoekscontext, (2) de inhoud van het protocol, (3) de politiek maatschappelijke zorgcontext, (4) de persoon en diens sociale- en zorgcontext, en (5) de mate van ervaren en benodigde competenties van de therapeut.

We concludeerden vanuit de resultaten dat een vlotte realisering van voorzieningen, een efficiënte multidisciplinaire samenwerking en meer mogelijkheden voor flexibele inzet van uren ergotherapie, de behandeling mogelijk kan verbeteren. Veel andere gevonden factoren geven informatie over alle aspecten die de ergotherapeut moet kunnen 'managen' (vereiste competenties) om een goede geïndividualiseerde behandeling te bieden.

## Hoofdstuk 7 – Algemene discussie en aanbevelingen

In dit hoofdstuk reflecteerde ik op de resultaten vanuit de verschillende OTiP deelstudies en geef op basis hiervan suggesties voor de praktijk en vervolgonderzoek.

De OTiP studie was, voor zover we weten, internationaal het eerste onderzoek dat de meerwaarde van ergotherapie bij de ziekte van Parkinson op een gedegen wetenschappelijke wijze heeft onderzocht. De studie toont aan dat de meerwaarde van de behandeling bestaat uit het verbeteren van ervaren uitvoering van dagelijkse activiteiten. Echter de studie geeft ook aan dat er noodzaak en ruimte is voor verbetering van de kwaliteit en effectiviteit van de behandeling.

Zo bleek uit de procesevaluatie dat aan een relatief groot deel van de doelen niet optimaal gewerkt was. Tevens gaven de ergotherapeuten aan dat het lastig was om voor patiënten in de begin of late fase van de ziekte mogelijkheden voor interventie te zien. Ze voelden zich - met name net na de start van het onderzoek- nog niet zo zeker in de uitvoering van alle stappen van het protocol. Hieruit trekken we onder andere de conclusie dat het belangrijk is om te blijven werken aan het vergroten van de competenties van de ergotherapeuten die parkinsonpatiënten behandelen. Naast ervaring en het bieden van gerichte nascholing, denken we dat persoonlijke feedback op professioneel handelen en het leren van elkaar zinvol is voor ergotherapeuten. Dit kan in de vorm van collegiale intervisie. Tevens kan gebruik gemaakt worden van informatie uit de kwaliteitsmonitor zoals die momenteel door ParkinsonNet wordt ingevoerd.

Een tweede punt dat in de OTiP studie naar voren kwam was de moeilijkheid om een tijdige en effectieve multidisciplinaire samenwerking te realiseren. Hoewel de organisatie van ParkinsonNet overleg tussen aangesloten zorgverleners potentieel mogelijk maakt, zijn niet altijd alle zorg- of dienstverleners die betrokken zijn bij een individuele patiënt, ParkinsonNet zorgverleners. Tevens leek de samenwerking nog teveel af te hangen van persoonlijke competenties van de zorgverleners. Dit benadrukt het belang van verdere aandacht voor verbetering van het inter-professioneel werken.

Een derde punt van de discussie was hoe ervoor gezorgd kan worden dat de afgesproken en aangeboden strategieën ook toegepast worden door de patiënten en mantelzorgers. Veel interventies binnen de ergotherapie betreffen een verandering in gedrag. Voorbeelden hiervan zijn activiteiten op een andere manier uitvoeren of activiteiten anders indelen in de dag. Om ervoor te zorgen dat deze gedragsverandering ook echt geïntegreerd wordt in het dagelijks leven, is een bepaalde intensiteit en periode van oefenen nodig. Er is echter nog maar weinig bewijs over wat die 'bepaalde' behandelintensiteit en -periode zou moeten zijn voor de strategieën die de ergotherapie inzet bij de ziekte van Parkinson. We hadden ook maar beperkt zicht op de mate waarin strategieën en adviezen echt werden opgevolgd door patiënten en mantelzorgers in de OTiP studie. Vanuit de dossiers (gerapporteerd door de therapeut) leek de mate van opvolging (adherentie) goed. In de focusgroepen gaven de

therapeuten wel aan dat adherentie wisselend was en dat het een belangrijke beïnvloedende factor was voor het succes van de behandeling. Vervolgonderzoek zou daarom nauwkeuriger in kaart moeten brengen welke strategieën ook echt opgevolgd en geïntegreerd worden in het dagelijks functioneren en hoe de adherentie bevorderd kan worden.

Een volgend punt in de discussie was de rol van de mantelzorger in de behandeling. De therapeuten vonden de betrokkenheid van de mantelzorger bij de behandeling van grote meerwaarde, maar er waren weinig mantelzorgers die ook een eigen hulpvraag hadden voor de ergotherapie. We deden de aanbeveling om in toekomstig onderzoek de hulpvragen van de mantelzorgers voor de diverse ziektestadia en de relevantie hiervan voor de ergotherapie nader te exploreren. Verder deden we de suggestie dat voor het adresseren van eigen hulpvragen van de mantelzorgers een groepsgericht programma misschien beter past dan een individueel traject.

We reflecteerden ook op de uitdaging om effecten te meten in een geïndividualiseerde interventie zoals de OTiP behandeling. We gebruikten in ons onderzoek de COPM en dit is een uitkomstmaat die de door de patiënt geprioriteerde problemen in activiteiten evalueert. Omdat individuele prioriteiten in de loop der tijd kunnen veranderen, kunnen behandeldoelen ook verschuiven en dan meet de COPM niet de effecten van de behandeling op die doelen. Verder onderzoek naar een uitkomstmaat die de effecten van een geïndividualiseerde ergotherapiebehandeling bij mensen met de ziekte van Parkinson kan vastleggen is gewenst.

Tot slot plaatsten we onze studies in het perspectief van de huidige transitie in de zorg. Tegenwoordig worden mensen geacht meer verantwoordelijkheid te nemen voor hun gezondheid en zo min mogelijk gebruik te maken van formele zorg. Tevens is er een verschuiving van ziekte management naar persoongerichte en individueel afgestemde zorg. We concludeerden dat de OTiP interventie heel goed past bij deze verschuivingen. We hebben echter een andere mening over het advies om specialisaties in de zorg te verminderen. Hoewel een heel aantal benodigde competenties generiek zijn, geven onze resultaten ook het belang aan van gespecialiseerde kennis en vaardigheden in het behandelen van mensen met de ziekte Parkinson. Op basis van onze resultaten hebben we onze aanbevelingen voor toekomstige ontwikkelingen als volgt samengevat:

- (1) Aanbevelingen direct toepasbaar binnen de huidige klinische praktijk:
  - Mensen met de ziekte van Parkinson die problemen ervaren in hun dagelijkse activiteiten of participatie dienen toegang te hebben tot een ergotherapeut met parkinsonexpertise.
  - Hoewel er verder onderzoek nodig is om aanbevelingen te kunnen aanscherpen, kunnen ergotherapeuten de richtlijn Ergotherapie bij de ziekte van Parkinson (2008) nog gebruiken als richtinggevend document voor de dagelijkse praktijk.

- Ergotherapeuten moeten voldoende aandacht besteden aan het ondersteunen van de patiënt en mantelzorger in het daadwerkelijk toepassen en integreren van afgesproken adviezen en strategieën.
  - Om een goede samenwerking in een geïntegreerd zorgnetwerk om de patiënt te realiseren, dienen zorgverleners hun kennis, vaardigheden en strategieën met elkaar te delen. Hiertoe kunnen ze gebruik maken van bestaande communicatiesystemen.
- (2) Aanbevelingen voor verdere implementatie van de OTiP interventie:
- Het in kaart brengen van de huidige werkwijze van ParkinsonNet ergotherapeuten is gewenst. Hierbij is het nodig te evalueren in hoeverre deze werkwijze aansluit bij de aanbevelingen van de richtlijn.
  - De belangrijkste determinanten voor implementatie van de OTiP interventie (interventie volgens de richtlijn) dienen geïdentificeerd te worden om vervolgens passende implementatie strategieën te ontwikkelen
  - Het wordt aanbevolen om de competenties van de ergotherapeuten actief te monitoren en trainingsmogelijkheden aan te bieden die passen bij verschillende expertise niveaus.
  - Alle belanghebbenden dienen actief betrokken te worden bij verder onderzoek naar implementatie van de interventie.
- (3) Aanbevelingen om de inhoud van de interventie verder te ontwikkelen en het bewijs voor ergotherapie bij de ziekte van Parkinson te vergroten:
- De mogelijkheden en de effecten van ergotherapie in de beginfase van de ziekte van Parkinson en in de late fase dienen nader in kaart gebracht te worden.
  - Een nadere exploratie is nodig van interventies die het best passen bij de hulpvraag van mantelzorgers in relatie tot 1) hun mogelijkheden en competenties om hun naaste met de ziekte van Parkinson te ondersteunen; en 2) het adresseren van hun eigen psychosociale hulpvraag.
  - Een beslismodel of middel dat het kiezen van passende aanpak bij een individuele situatie gericht ondersteunt dient ontwikkeld te worden.
  - Er is nader onderzoek nodig om een enkele of samengestelde uitkomstmaat te ontwikkelen die de diverse effecten van geïndividualiseerde ergotherapie bij mensen met de ziekte van Parkinson kan meten.
  - In bovengenoemde onderzoeken dienen mensen met de ziekte van Parkinson en hun naasten zoveel mogelijk als partners in het onderzoek betrokken te worden en niet alleen als deelnemers.



**Dankwoord**

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**List of publications**

**RIHS PhD portfolio**

**Dissertations of the Parkinson Centre**



## Dankwoord

Het onderwerp ergotherapie bij Parkinson ligt me na aan het hart en onderzoek doen vind ik leuk, dus dit promotietraject ben ik vol enthousiasme aangegaan. Maar al met al is het ook veel geweest en bij tijden lastig. Daarom aan iedereen die mij direct of indirect geïnspireerd, geholpen en gesteund hebben om de OTiP studie uit te voeren en dit promotietraject te volbrengen wil ik zeggen: Dank jullie wel! Ik had deze 'klus' onmogelijk alleen kunnen klaren. Een aantal personen en groepen wil ik in het bijzonder noemen:

Ten eerst mijn begeleidingsteam. Wat heb ik het getroffen met jullie! Ria, je was net begonnen als hoogleraar paramedische wetenschappen toen ik met mijn traject begon. Als mijn 1<sup>e</sup> promotor ben je tijdens het gehele proces intensief betrokken geweest. De vele inhoudelijke discussies met jou leverden altijd nieuwe inzichten op. Je was snel, grondig en prettig kritisch in je feedback op mijn stukken, en je had altijd oog voor mijn persoonlijk welzijn. Dit alles heb ik erg gewaardeerd. Maud, jij was hier ook deelgenoot van. Samen met Ria vormde je mijn kernteam. In Ergoland ben je *het* grote voorbeeld van een gedegen en succesvolle ergotherapie onderzoeker. Jouw kennis en ervaringen hebben mij erg geïnspireerd. Wat gezellig was het ook om samen met jou naar internationale ergotherapie congressen te gaan. Ik heb speciale herinneringen aan onze tijd in Japan.

Bas en Marten, het andere duo van mijn begeleidingsteam. De passie die jullie hebben voor de ziekte van Parkinson en parkinsonzorg is echt aanstekelijk. Ik ben dan ook heel blij deel uit te maken van jullie team en missie.

Bas, jij bent een groot fan van heel veel...en zo ook van de ergotherapie. Geweldig dat het eerste wetenschappelijk bewijs van de meerwaarde van de ergotherapie in de multidisciplinaire parkinsonzorg er nu is. Naast 'Bloem's writing tips' ben ik je erg dankbaar voor de finishing touch in de laatste versies van veel van mijn schrijfwerk. Je weet als geen ander een boodschap goed te formuleren en uit te dragen.

Marten, door een verschuiving van rol was je tijdens de schrijffase meer op de achtergrond, maar je was onmisbaar voor de basis van het hele traject. Je bent intensief betrokken geweest bij de ontwikkeling van de richtlijn, de subsidieaanvragen en de opzet van het onderzoek. Dank voor je hulp en je vertrouwen in mij. Mijn promotiedag is tevens jouw verjaardag! Dubbel feest.

Lieve Yvonne, je was een onderzoeksassistent waar ik op kon rekenen en je hebt me met alles enorm geholpen. Je kwam in 2009 bij me met de vraag of je een onderzoeksstage kon doen voor je opleiding gezondheidswetenschappen. Dat was een perfecte timing en match. Je hebt goed werk geleverd met de diepte-interviews voor de pilot studie. Daarna ging je aan de slag als onderzoeksassistent bij de grote OTiP studie. Je was zo flexibel en hebt zoveel werk verzet. Leuk dat je nu je eigen onderzoek

hebt en we weer gezellig het kantoor delen. En geweldig dat je op de grote dag mijn paranimf bent.

Bart, je was de langste tijd mijn 'OTiP-secretaresse'. Eindeloos brieven vouwen, belletjes plegen en data invoeren. Eigenlijk helemaal niet je ding, maar je deed het toch: rustig, nauwkeurig en met een dosis humor.

Ook alle assistenten, studenten en ergotherapeuten die naast Bart en Yvonne hebben bijgedragen aan de dataverzameling, data invoer of het scoren van PRPP observaties wil ik nogmaals hartelijk danken.

Alle patiënten, mantelzorgers en ergotherapeuten die hebben deelgenomen aan de studies wil ik ook zeker bedanken. Zonder jullie zou ik geen data hebben. Het heeft de nodige tijd en inzet gevraagd om deel te nemen en geëvalueerd te worden. Ik hoop dat het uiteindelijk voor ieder van jullie de moeite waard was.

Jan Hendriks, mijn grote dank voor je statistische ondersteuning. Je hebt veel berekeningen voor me gedaan en ik heb veel van je geleerd. Ik denk nog steeds aan je als ik weer tabellen moet opmaken, of anderen tips geef hierover: 'Jan's manier'.

I am also very grateful towards the members of my doctoral thesis committee: professor Marcel Olde Rickert, professor Teus van Laar and professor Tanya Packer. Thank you for taking the time to review my manuscript. Tanya, your expertise in the field of occupational therapy and self-management is very inspiring. I hope we can collaborate in the future in my endeavors to continue improving the quality of occupational therapy services in the field of Parkinson's disease.

Dan wil ik mijn directe collega's als groep noemen. Ik was al ingebed in twee geweldige teams toen ik aan mijn promotie begon: revalidatie (met name de sectie ergotherapie) en ParkinsonNet. De samenstelling van de teams is in de loop der jaren wel wat veranderd, maar de gezelligheid, gedrevenheid, respect en persoonlijke aandacht blijven kenmerkend. Dank lieve collega's, het is heel prettig om met jullie samen te werken! We delen allen de drijfveer om de zorg voor de patiënt continue te willen verbeteren.

Qua ruimte was het passen en meten, maar toch was het goed vertoeven in 'het holletje' dankzij mijn mede-holbewoners (of ex-penthousers). Edith, inmiddels ben je toegewijd hoofd van onze sectie, maar met je eigen kantoor naast het holletje, krijg je toch nog van alles mee. Als fijne collega ergotherapeut, onderzoeker, en mijn leidinggevende, heb je me altijd goed gesteund in de afgelopen jaren. Dank voor je

luisterend oor, je begrip en bereidheid om altijd te zoeken naar mogelijkheden. Joyce, jij bent al weer eventjes uitgevlogen van het holletje. Je zorgde altijd voor levendigheid en prettige afleiding als ik weer eens te veel 'onverstoorbaar' aan mijn scherm geplakt zat. Je was een goede spiegel van mijn eigenaardigheden. Ik mis je aanwezigheid in het holletje.

Hanneke, je was niet direct betrokken bij mijn onderzoek, maar je bent al zo lang zo'n fijn ParkinsonNet maatje. Jouw professionaliteit, rust, en persoonlijke aandacht heeft me veel geholpen in het ontwikkelen van de richtlijn en de Expert werkzaamheden binnen ParkinsonNet. Leuk dat je deze dag ook naast (achter) me wil staan als mijn paranimf.

Dan mijn 'Femina' vriendinnen Ellen, Selma, Petra en Dorien. We kennen elkaar van de opleiding in Hoensbroek en hebben door de jaren heen contact gehouden. We houden die vriendinnen dagjes uit er zeker in! Het is altijd weer gezellig en vertrouwd. Selma, leuk dat je ook betrokken was als ParkinsonNet ergotherapeut in zowel de pilot studie als de grote studie. Ellen, vooral jou heb ik door de jaren regelmatig 'belast' met de dilemma's die ik tegenkwam in het balanceren van werk en privé. Dank voor je luisterend oor en altijd wijze raad.

Lieve papa en mama, van alles wat jullie me van jongs af aan hebben meegegeven, heb ik hard werken, volhouden en optimistisch blijven zeker nodig gehad om dit traject te volbrengen! Dank voor jullie praktische en emotionele steun en jullie vertrouwen in de beslissingen die ik neem. Angélique, Miriam, en William, we volgen inmiddels allemaal al lang ons eigen pad. Maar ik geniet steeds van de momenten dat we elkaar met onze gezinnen spreken en zien. Dank voor jullie interesse en betrokkenheid bij mij, jullie verstrooide zus(je).

'Last but not least' mijn gezin en thuis; mijn lieve Juan, Eloine, en Carmen. Jullie moesten geduld hebben met de vele uurtjes die ik extra achter de computer spendeerde. Eloine en Carmen, jullie weten haast niet beter. Eerst de richtlijnontwikkeling en toen dit promotietraject naast andere werkzaamheden. Eloine, jouw gezellige gekwebbel en Carmen, jouw lieve knuffels en grapjes geven me altijd veel plezier en energie. Ik denk niet dat ik ooit een rustige baan zal hebben, maar ik kijk ernaar uit dat ik de weekenden weer onbezorgd vrij kan zijn en tijd heb voor leuke dingen met z'n viertjes. Ook voor ons Juan.

En nu eerst samen genieten van deze bijzondere dag.

## About the author



*De Venster Fotografie*

Ingrid Sturkenboom was born on 5<sup>th</sup> October 1969 in Houten, the Netherlands. After finishing pre-university secondary education, she followed from 1988 to 1992 a bachelor-level occupational therapy training at the Hogeschool Heerlen in Hoensbroek. After her graduation, she moved to England, where she worked as an occupational therapist in different hospital settings. In 1994 she started a one-year 'round-the-world-backpacking trip' during which she also worked as an occupational therapist in New Zealand for a few months. Upon return, she worked on a temporary contract in Nijmegen, The Netherlands, but then decided to return to England. From 1996 to 2000 she worked as a senior occupational therapist mainly in Liverpool in the field of neurological rehabilitation. Next, she was recruited by an English development agency to work for two years as a volunteer trainer at the Kilimanjaro Christian Medical College in Moshi, Tanzania. There she helped with the set up and running of the first Tanzanian school for occupational therapy.

Interested in the impact of culture on perceptions of health, disability and healthcare, she started in 2003 the international Amsterdam Master's in Medical Anthropology at the University of Amsterdam. She graduated cum laude in 2004.

In 2005 Ingrid was appointed as an occupational therapist at Radboud university medical center, with a special focus on Parkinson's disease. Since 2007 she has been the expert/coordinator for occupational therapy within the national project team of ParkinsonNet. She is first author of the Dutch guidelines for occupational therapy in Parkinson's Disease (2008) and started in 2009 her PhD research on the effectiveness of occupational therapy in Parkinson's disease: the OTiP study. In the following years to date she combined her PhD research, (international) training of professionals, participation in other projects/committees, and patient care.

Ingrid is married with Juan Carlos Gonzalez Pelayo and together with their two daughters, Eloine (9) and Carmen (7), they live in Meteren.

## Over de auteur

Ingrid Sturkenboom is geboren op 5 oktober 1969 te Houten en groeide voornamelijk op in Oosterwolde (Fr). Na afronding van de middelbare school (Atheneum-B), volgde ze van 1988 tot 1992 een bachelor opleiding Ergotherapie aan de Hogeschool Heerlen te Hoensbroek. In 1992 begon ze haar carrière als ergotherapeut in Engeland in verschillende ziekenhuizen en centra. In 1994 reisde ze een jaar 'rond de wereld' en werkte in die periode een paar maanden als ergotherapeut in Nieuw Zeeland. Bij terugkomst werkte ze tijdelijk in de St Maartenkliniek in Nijmegen. In 1996 besloot ze terug te keren naar Engeland en werkte daar tot eind 2000, voornamelijk in Liverpool in het werkveld van neurologische revalidatie. Vervolgens werkte ze als vrijwillige trainer voor een Engelse ontwikkelingsorganisatie bij Kilimanjaro Christian Medical College, in Moshi, Tanzania. Daar hielp ze met het verder opzetten van de eerste Tanzaniaanse ergotherapie opleiding en gaf les.

Geïnteresseerd in de invloed van cultuur op de perceptie van gezondheid, handicap en zorg, startte ze 2003 de internationale 'Amsterdam Master's in Medical Anthropology' opleiding aan de Universiteit van Amsterdam. Ze studeerde in 2004 cum laude af.

In 2005 werd Ingrid aangenomen als ergotherapeut bij het Radboudumc, met speciaal aandachtsgebied de ziekte van Parkinson. Sinds 2007 is ze expert/coördinator ergotherapie van het nationale ParkinsonNet projectteam. Ze is eerste auteur van de Nederlandse richtlijn Ergotherapie bij de ziekte van Parkinson (2008) en startte in 2009 haar promotieonderzoek naar de effectiviteit van ergotherapie bij de ziekte van Parkinson: de OTiP studie. Sindsdien combineert ze haar promotieonderzoek, (internationale) training van zorgverleners, andere projecten/commissies, en patiënten-zorg.

Ingrid is getrouwd met Juan Carlos Gonzalez Pelayo en samen met hun twee dochters, Eloine (2006) and Carmen (2008), wonen ze in Meteren in de Betuwe.

## RIHS PhD Portfolio

Institute for Health Sciences  
Radboudumc

Name PhD student: IHWM Sturkenboom  
Department: Rehabilitation  
Graduate School: Radboud Institute for Health Sciences

PhD period: 01-03-2009 to 11-02-2016  
Promotors: Prof.dr. MWG Nijhuis-van der Sande, prof dr. BR Bloem  
Co-promotors: Dr. MJL Graff, dr. M Munneke

	Year(s)	ECTS 1
<b>TRAINING ACTIVITIES (related to PhD trajectory)</b>		
<b>a) Courses &amp; Workshops</b>		
- Time management voor promovendi	2009	0.3
- BROK (Certificate Good Clinical Practice)	2010	1.0
- Biometrics (extended version)	2010-2011	3.0
- KWALON Training Focusgroepen	2011	0.8
- RUNMC Workshop: Networking	2011	0.1
- PhD Workshop on Statistics	2012	0.1
- The art of presenting science	2013	1.5
- Academic Writing	2013-2014	3.0
- Re-registration BROK	2014	0.1
- Masterclass 'Waar gaat hij nou naartoe'?	2015	0.2
<b>b) Seminars &amp; lectures</b>		
- NWO Talentendag, Utrecht	2010	0.3
- NCEBP (RIHS) thema bijeenkomst, Evaluatie van complexe interventies	2010	0.1
<b>c) Symposia &amp; congresses</b>		
- NCEBP (RIHS) Symposium	2009, 2012	0.5
- World Parkinson congress, Glasgow, Scotland; 2 poster presentations	2010	1.8
- Jaarcongres Ergotherapie, Utrecht; 2 poster presentations	2010	0.5
- ParkinsonNet Jaarcongres; oral presentation	2010	0.5
- Symposium Ouderen 'n update', Utrecht; oral presentation	2011	0.5
- 9th COTEC Congress of Occupational Therapy, Stockholm, Sweden; 2 oral presentations	2012	1.3
- World Parkinson congress, Montreal, Canada; 2 poster presentations with 1 guided poster tour and organization of international OT meeting.	2013	2.2
- ParkinsonNet Jaarcongres; oral presentation	2013	0.5
- Jaarcongres Ergotherapie, Utrecht; oral and poster presentation	2013	0.8
- World Federation of OT Congress, Yokohama, Japan; oral presentation	2014	1.3
- Symposium Ergotherapie en ouderen, oral presentation	2014	0.5
- Jaarcongres Ergotherapie, Apeldoorn, poster presentation	2014	0.5
- ParkinsonNet Jaarcongres, oral presentation	2014	0.5

-	Joint HKMDS and HKOTA Symposium and Workshop on Parkinson's disease, Hongkong, China; keynote lecture	2015	0.8
d)	Other		
-	Biweekly ParC researchers' lunch meeting, Radboudumc	2010-2012	3.0
-	Meetings NEON (Netwerk voor Ergotherapie Onderzoekers in de regio Nijmegen)- 4 times a year	2009-to date	2.0
TEACHING ACTIVITIES			
e)	Lecturing (indirectly related to PhD research)		
-	Lecturing in post graduate ParkinsonNet courses and study days (national)	2007-to date	NA
-	Lecturing in post graduate ParkinsonNet courses (international)	2015-to date	NA
-	Joint HKMDS and HKOTA Symposium and Workshop on Parkinson's disease, Hongkong, China; 6 workshops	2015	NA
-	Yearly lecture in Module V Biomedical students	2008-to date	0.5
f)	Supervision of internships/other (related to role as researcher)		
-	Supervision internship MSc student Gezondheidswetenschappen	2009-2010	1.5
-	Review/rate 4 research projects BSc OT students	2008-2014	0.5
-	Research advisor student MSc Neurorehabilitation	2014-2015	0.5
-	External Data monitor for Energetic study	2014-to date	0.3
-	Review scientific publication	2015	0.1
<b>Total ECTS</b>			<b>30.8</b>



## Peer reviewed publications (\* listed in this thesis)

2015	<b>*Sturkenboom IHWM</b> , Nijhuis-van der Sanden MWG, Graff MJL. A process evaluation of a home-based occupational therapy intervention for Parkinson's patients and their caregivers: performed alongside a randomized controlled trial. <i>Clinical Rehabilitation</i> . Accepted.
	Poerbodipoero SJ, <b>Sturkenboom IH</b> , van Hartingsveldt M, Nijhuis-van der Sanden MWG, Graff MJ. The Construct Validity of the Dutch Version of the Activity Card Sort. <i>Disability and Rehabilitation</i> . Accepted.
	<b>*Sturkenboom IHWM</b> , Hendriks JCM, Graff MJL, Adang EM, Munneke M, Nijhuis-van der Sanden MW, Bloem BR. Economic Evaluation of occupational therapy in Parkinson's disease: a randomized controlled trial. <i>Movement Disorders</i> 2015; 30 (8):1059-1067.
2014	<b>*Sturkenboom IHWM</b> , Graff MJL, Hendriks JCM, Veenhuizen Y, Munneke M, Bloem BR, Nijhuis-van der Sanden MW, for the OTiP study group. Efficacy of occupational therapy for patients with Parkinson's disease: a randomised controlled trial. <i>The Lancet Neurology</i> 2014; 13(6):557-66.
2013	<b>*Sturkenboom IH</b> , Graff MJ, Borm GF, Adang EM, Nijhuis-van der Sanden MW, Bloem BR, Munneke M. Effectiveness of occupational therapy in Parkinson's disease: study protocol for a randomized controlled trial. <i>Trials</i> 2013; 14:34.
	<b>*Sturkenboom IH</b> , Graff MJ, Borm GF, Veenhuizen Y, Bloem BR, Munneke M, Nijhuis-van der Sanden MW. The impact of occupational therapy in Parkinson's disease: a randomized controlled feasibility study. <i>Clinical rehabilitation</i> 2013; 27(2):99-112.
2012	Steultjens E, <b>Sturkenboom I</b> . Richtlijnen Lust of Last? <i>Wetenschappelijk Tijdschrift voor Ergotherapie</i> 2012; 5(1):52-56.
2010	<b>Sturkenboom I</b> , Graff MJL. Ergotherapie aan huis. Een beschouwing vanuit onderzoek en praktijk. <i>Wetenschappelijk Tijdschrift voor Ergotherapie</i> 2010; 3(4):20-22.
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ISBN 978-94-6259-989-5

