New models of care

Esther H.A. van den Bogaart

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focusing on substitution of hospital care with primary care.
Trend setting or trend breaking?

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PROEFSCHRIFT

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Contents

01

General introduction

Our life expectancy has risen by nine years since the mid-twentieth century, half of which is due to improved healthcare [1]. As healthcare costs are strongly age dependent, these gains in longevity will lead to higher healthcare expenditure [2, 3]. With the 'silver tsunami' in the offing, the sustainability of healthcare systems worldwide is threatened [4]. Along with the ageing population, increasing rates of chronic and complex diseases and new expensive medical technologies and medicines are putting our healthcare systems under pressure [5, 6]. The additional lack of value-consciousness and high expectations among healthcare consumers, health workforce shortages, and inefficiencies in healthcare delivery further raise concerns about guaranteeing accessibility and affordability of healthcare in the near future [7-9].

In most countries affiliated with the Organisation for Economic Cooperation and Development (OECD), health spending rises faster than economic growth [10]. It is estimated that health spending per capita will further grow at an average annual rate of 2.7% and will reach on average 10.2% (ranging from 4.6% to 20.2%) of the gross domestic product (GDP) by 2030. However, despite these growing healthcare expenditures, a proportionally increase in health status is lacking, indicating that much of the spending is most likely unnecessary [11].

FRAGMENTATION

Despite the fact that care provision can be regarded as collective work, healthcare organisations have a tendency to work autonomously in 'distinct silos' [12, 13]. This fragmentation of care is often pointed out as an underlying problem of unnecessary care and high expenses in healthcare [14, 15]. Fragmentation can be found in many countries in which health services are strictly divided into primary and secondary care and refers to the misalignment of incentives and the lack of coordination, leading to an inefficient allocation of resources and negatively impacting quality, costs and outcomes [16, 17]. As a result, patients are often transferred to other healthcare settings and organisations without complete information about their condition, medical history, previously provided services or prescribed medications. This entails risks, waiting times and delays for the patient and unnecessary and/or duplicate tests and treatments causing additional costs [18].

Fragmentation is the result of the rapid expansion of medical knowledge and the increasing specialisation of healthcare providers [4, 19]. Due to the process of specialisation, healthcare systems were divided into 'pigeon-holes' with their own

specialist fields, interests, positions of power and funding [20]. By dividing doctors into specialties and subspecialties with increasing technical skills, it was assumed that health outcomes would improve [21]. However, despite specialisation is inevitable and useful, 'too much' specialisation may lead to a lack of care coordination and continuity which has adverse impacts on patient outcomes, care experience and healthcare costs [22, 23]. In addition, the fragmentation of healthcare budgets further increases the challenges for efficient and effective care since separate budgets for primary and secondary care are obstructing care coordination and professional collaboration and communication and facilitating overuse of care [14, 24, 25].

CARE COORDINATION

Coordinating the services of different care providers along the continuum of care is essential to overcome fragmentation problems [26]. With coordination of care as one of its core functions, primary care plays an important role in this process [27, 28]. In many healthcare systems, primary care serves as a patient's entry point to the healthcare system (gatekeepers function) with the general practitioner (GP) integrating all aspects of care when patients need to be seen elsewhere and guiding patients to the most appropriate providers [18]. Information exchange between GPs and medical specialists is an important part of this process. However, communication between healthcare providers can be optimised since referrals from GPs to specialists often contain insufficient information, and specialists reports back to the GP are often late and inadequate [29, 30]. This can result in unnecessary referrals and expenses, and suboptimal quality and experience of care [31].

INTEGRATED CARE

In response to fragmentation and care coordination problems in healthcare and to transform healthcare delivery, efforts have increasingly focused on integrated care [32]. Integrated care has become a central part of policy initiatives worldwide in order to enhance the sustainability and affordability of healthcare systems [33]. According to Singer et al. [32] integration refers to 'a set of organisational and social features and courses of action or activities requiring unification that may exist both within and between organisations'. Integration may occur along four different dimensions: functional, organisational, professional, and clinical [34]. Functional integration aims to coordinate key support functions and activities, and refers to formal protocols and policies [32, 35, 36]. Organisational integration refers to formally bringing together

of healthcare organisations through contractual arrangements and alliances [36]. Professional integration focuses on formal collaboration between (multidisciplinary) healthcare professionals, both within and between institutions. Finally, clinical integration refers to organisational activities designed to coordinate patient care services across people, functions, activities, and operating units over time to maximise the value of services delivered to patients, for example through the use of shared guidelines and protocols [35, 36].

In addition to the division by dimension, integration can be considered from the level at which integration takes place [18]. Horizontal integration refers to the linking of care, services or functions at organisations of the same type (for example mergers of healthcare organisations) [32]. Vertical integration refers to the linking of different levels of care, services, or functions among organisations of different types, within and outside healthcare (for example integrating primary and secondary care) [32, 37].

Furthermore, integrated care can be classified according to its intensity, covering a spectrum from 'no or partial integration' to 'full integration' [38]. Partial integration refers to collaboration between organisations which retain their own service responsibility and funding criteria while full integration involves the process of integrating into a new organisational model responsible for the full continuum of care [34].

PATIENT-CENTRED MEDICINE

To deliver integrated care, it is no longer enough to provide better services solely based on improving skills, clinical procedures and high-technology [18, 39]. Instead of illness-oriented healthcare systems focusing on a single clinical picture (in both the delivery and financing of care), a shift to whole-person care is needed, [40, 41]. Patient-centred medicine aims to improve patients' health outcomes in everyday clinical practice, with taking into account patients' preferences, objectives and values, as well as their available resources. Or as stated by William Osler [42]: 'It is more important to know what sort of patient has a disease than what sort of disease a patient has'. To achieve a more patient-centred approach, structural changes in healthcare systems are needed to overcome divergent and fragmented priorities, metrics, outcomes and budgets among providers working across different settings and organisations [43].

BARRIERS OF INTEGRATED CARE

Despite the argumentation that healthcare will become increasingly fragmented, inefficient and unsustainable when a patient-centred and integrated approach is lacking, the integration of primary, secondary and tertiary care in many European countries is still weak [18, 44]. As mentioned by Kozlowska et al. [43], barriers to successful implementation of integrated care, including among others, are a lack of commitment by organisations involved, conflicting interests, insufficient resources, and resistance to change. Furthermore, there is no 'one size fits all' approach in which different parts of healthcare can seek to achieve more integrated care [43]. As a result, efforts on care integration have targeted different objectives at varying levels within and across healthcare systems [32]. Examples of ways to integrate care are: care pathways, case or disease management, re- or co-location like community-based or specialist outreach clinics, multidisciplinary teams, and shared guidelines and/or protocols [43]. Although these initiatives vary widely in structure and style, they all aim to improve health outcomes while containing overall healthcare costs [45, 46].

TRIPLE AIM

According to the Triple Aim framework, to achieve successful care integration, three aims should be linked: (1) reduced care costs should coincide with (2) improved population health and (3) improved patient experiences (see Figure 1) [47]. To realise these aims, organisations need to collaborate, change their culture and start thinking outside the boundaries of their own organisation [48]. Furthermore, since all Triple Aim dimensions are interdependent, changes pursuing one dimension can affect the other two, both positively and negatively. Therefore, a balanced and concurrent pursuit of all dimensions to ensure affordable high-quality care delivery is needed [49].

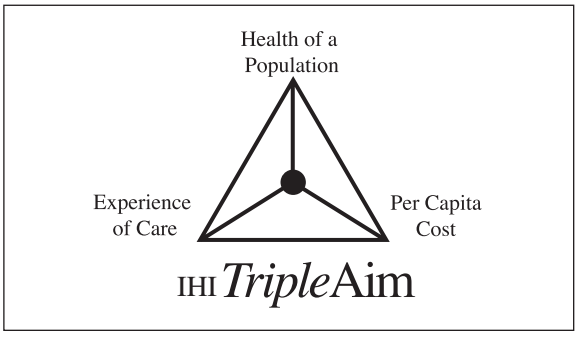

Figure 1 The Triple Aim according to the Institute for Healthcare Improvement (IHI) [72]

Since its introduction in 2008, the Triple Aim framework is increasingly used to design and assess initiatives in healthcare. The framework can be used to guide organisations in achieving their goals. According to Berwick et al. [47], there are three preconditions for a successful implementation of the framework. First, an 'integrator' is needed who is able to focus and coordinate services within the healthcare system. The integrator needs to ensure a continuum of care by making connections between stakeholders and resources. Second, the population of concern needed to be defined. Finally, budget constraints and clear policy levers must be recognised that claim principles of health equity.

NEW MODELS OF CARE

To experiment with different forms of care integration, health authorities worldwide are implementing new models of care at local level in sites (so-called pilot sites, pioneer sites, or vanguards) that act as blueprints and inspiration to the rest of the healthcare system [50]. Examples include the pioneers and vanguards in the National Health Service (NHS) in the United Kingdom, where multispecialty community providers focusing on moving specialist care out of hospitals into the community and acute hospital services are joining up with GPs, community, mental health and social care services. Similar examples can be found in other European countries, North America, Australia, and New Zealand [51-57].

Within the same country, new models of care may take different forms as they are tailored to the local needs and are dependent of and interact with local context [58-60].Therefore, innovation is driven by local leaders from multiple-organisations who collaborate and are responsible to improve access and quality of care for their local populations [61].

SUBSTITUTION

One of the strategies to provide more integrated care as well as increasing efficiency of care is shifting specialised medical care from hospital based outpatient care to primary care without changing the people who deliver the service [62]. This refers to relocation or substitution of care. According to Warner [63] substitution can be defined as 'the continual regrouping of resources across and within care settings, to exploit the best and least costly solutions in the face of changing needs and demands'. Substitution is driven by the broad consensus that (a part of) care should

be shifted from hospital to community in the coming years to reduce costs [64].

With hospitals worldwide typically consuming between 40 and 80% of the total healthcare expenditure, secondary care services have higher costs in comparison with primary care [65, 66]. Furthermore, secondary care is characterised by access problems, including long waiting times for specialised medical care [67, 68]. As increasing the capacity and availability of hospital consultations does not automatically lead to better access for all patient groups, expanding secondary care services is not the solution to meet the increasing demand [68]. Therefore, many countries have focused their reform efforts on strengthening primary care since it is characterised as essential, affordable and accessible care addressing the needs of all patients in the community and integrating care, prevention, promotion and education [69, 70]. By focusing on substitution of specialist medical services in the community, primary care is strengthened by embedding specialist medical knowledge [71]. Furthermore, by focusing on substitution, unnecessary referrals to secondary care can be prevented and/or avoided and care can be delivered closer to the patient [46].

DUTCH PIONEER SITES

As in many other countries, improving the financial sustainability of the healthcare system is high on the Dutch political agenda [72]. Health expenditure has risen sharply since 2008, making the Netherlands one of the top health spenders worldwide [73]. Therefore, regional innovation initiatives are designated as pioneer sites in a nationwide effort to achieve 'better healthcare at lower cost' [48]. The initiatives implemented in nine pioneer sites across the country (see Figure 2) are in line with the 'the right care in the right place' movement initiated by the Dutch Ministry of Health, Welfare and Sport. This movement is focusing on avoiding expensive care, moving care delivery closer to people's homes and replacing traditional care delivery with new forms such as e-health [20]. Moreover, the initiatives are implemented by different collaborating regional organisations (like healthcare providers, health insurers, patient and public representatives and municipalities) to improve the health of the population and quality of care, and to limit the growth of healthcare expenditure (Triple Aim) [74].

Two of these pioneer sites are located in the province of Limburg, in the south of the Netherlands: 'Blue Care' (in Dutch: 'Blauwe Zorg') and 'MyCare' (in Dutch: 'MijnZorg'). One of the health insurance companies later added a third pioneer site in this region,

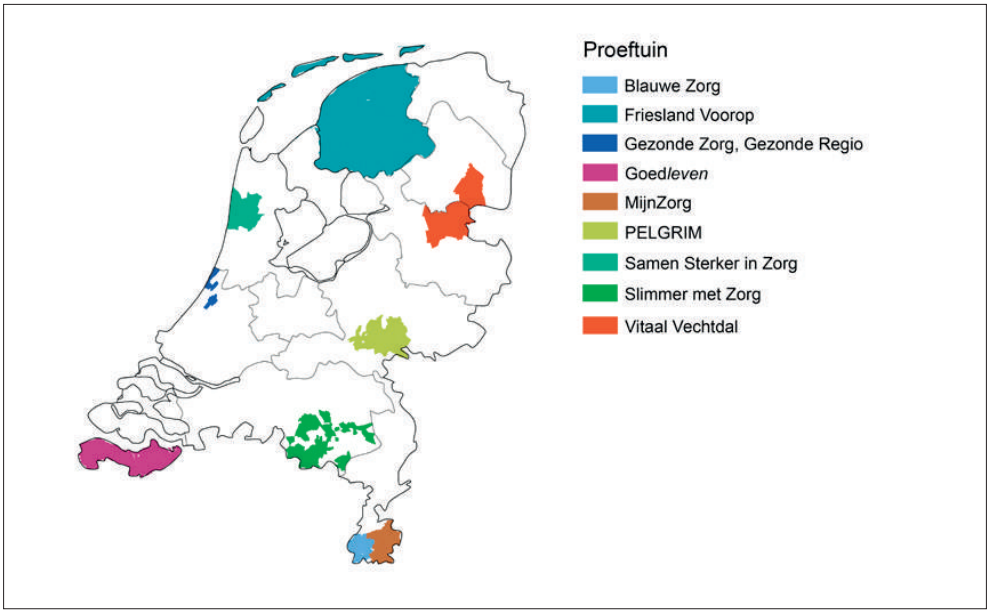

Figure 2 The nine Dutch pioneer sites

named 'Differently Better' (in Dutch: 'Anders Beter'). This pioneer site is located just above the other two pioneer sites in the south of the Netherlands. In these three pioneer sites, substitution of care is one of the strategies to control rising healthcare costs.

In particular, the south of the province of Limburg is characterised by relatively high use of care and an unhealthy population with a shorter life expectancy compared to the rest of the Dutch inhabitants [75]. The high urgency in this region is making the pioneer sites all the more interesting.

AIM OF THIS DISSERTATION

The main focus of this dissertation is on a new model of care named Primary Care Plus (PC+) implemented in the Dutch pioneer site 'Blue Care' located in the Maastricht-Heuvelland region in 2013. PC+ is a substitution initiative, focusing on medical specialists providing face-to-face consultations in a primary care setting. PC+ is a form of vertical integration in which primary and secondary care are linked through a joint venture (organisational integration). Interventions related to PC+ described in previous literature are for example joint consultations [76, 77] and outreach services or clinics [78-82].

The PC+ initiative is a collaboration between the primary care organisation 'Care in Development' (in Dutch 'Zorg in Ontwikkeling'), the Maastricht University Medical Centre+ (Maastricht UMC+), the health insurance company VGZ, and the patient representative foundation 'Burgerkracht Limburg'. After the pilot phase (2013-2014) in which medical specialists performed consultations in various GP practices, two independent PC+ centres were established in 2014 to overcome problems with inefficient planning of consultation hours, low patient numbers and overuse of care [83]. The independent PC+ centres are located in a neutral primary care setting outside the hospital premises to which GPs can refer non-acute and low-complex patients. In PC+, patients are seen by a (senior) medical specialist during a maximum of two consultations while the GP remains responsible for the patient. Following PC+, patients are referred back to their GP with a treatment advice, or, if necessary, they are referred to hospital care for further diagnosis and/or treatment.

Additionally, this dissertation also includes the findings of the care pathway 'Better exercise in osteoarthritis' implemented in pioneer site 'Differently Better' located in the Western Mining District. The pathway aims to aims to treat patients with knee or hip osteoarthritis according to a stepped care approach. Although this pathway is implemented in a different pioneer site and differs from the PC+ intervention, it also focuses on reducing unnecessary care (i.e. GP diagnostic imaging requests) and avoiding or delaying hospital visits (i.e. GP referrals to orthopaedic surgeons). Designed based on guidelines, the pathway is focusing on clinical integration between primary and secondary care (vertical integration). Adding the research into this pathway to this dissertation adds insight into varying possibilities for substitution.

The aim of this dissertation is to study the effect of both interventions on the referral behaviour to secondary care and/or for diagnostic imaging. Furthermore, this dissertation attempts to identify the influence of predictive characteristics on the decision whether to refer to secondary care. Finally, the effect on patients' health-related quality of life and the patients' experienced quality of care, the costs-effectiveness, and the volume of care on a regional level was explored. Based on these aims, two objectives have been formulated:

To assess the influence of the implementation of PC+ and the care pathway 'Better exercise in osteoarthritis' on the referral behaviour from primary to secondary care and the request of diagnostic imaging, and to determine the influence of patient and consultation characteristics on this referral decision.

To determine how PC+ effects the outcomes of the Triple Aim and regional healthcare volumes and which economic evaluation approach can be used best to decide whether to invest in PC+.

OUTLINE

Based on these two objectives, this dissertation consists of two main parts. **Part I** includes chapters 2, 3 and 4 and answers the first research question. The first two chapters give insights in the referral decision following PC+ in the 'Blue Care' region (i.e., referral back to the GP or referral to outpatient hospital care), but are focusing on a different medical specialty. **Chapter 2** is focusing on dermatology care and describes the influence of predictive patient and consultation characteristics on the referral decision following dermatology care in PC+. **Chapter 3** describes findings regarding the referral decision following orthopaedic care in PC+ and the influence of the availability of diagnostic tests in PC+ on this referral decision. The results of both chapters can be used as input for the optimisation of the PC+ process. **Chapter 4** is focusing on a stepped-care approach, in the shape of the care pathway 'Better exercise in osteoarthritis', which was implemented in the Western Mining District of Limburg. The focus of this chapter is on the effect of the pathway on GP diagnostic imaging requests and GP referrals to orthopaedic surgeons for hip and knee osteoarthritis and to what extent the pathway is applied in.

Part II of this dissertation includes chapter 5, 6 and 7 and only includes findings of the 'Blue Care' region by focusing on the effect of PC+ on the patient level and regional level. **Chapter 5** describes the patient-reported health-related quality of life and the experienced quality of care from patients referred to PC+ and compares this to the patient-reported outcomes from patients referred to hospital based outpatient care using propensity score matching. Whether PC+ is cost-effective compared to hospital based outpatient care is described in **chapter 6**. In this chapter, a side-by-side application of the cost-utility analysis and multi-criteria decision analysis is conducted to investigate the applicability and suitability of both methods related to the economic evaluation of new models of care, like PC+. The aim of this chapter is to investigate whether the adoption of multi-criteria decision analysis, instead of traditional cost-utility analysis, alters the decision of investing in PC+. **Chapter 7** gives insights whether PC+ actually succeeds in shifting outpatient hospital care to the primary care setting and therefore results in substitution of care on a regional level by investigating the effect of PC+ on the regional healthcare volumes.

Finally, the general discussion describes the main findings as well as the theoretical and methodological reflections on these findings. Furthermore, it addresses the outcomes on daily practice by focusing on the implications for future policy, practice and research.

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02

Reorganising dermatology care

Predictors of the substitution of secondary care with primary care

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ABSTRACT

Background: The substitution of healthcare is a way to control rising healthcare costs. The Primary Care Plus (PC+) intervention of the Dutch 'Blue Care' pioneer site aims to achieve this feat by facilitating consultations with medical specialists in the primary care setting. One of the specialties involved is dermatology. This study explores referral decisions following dermatology care in PC+ and the influence of predictive patient and consultation characteristics on this decision.

Methods: This retrospective study used clinical data of patients who received dermatology care in PC+ between January 2015 and March 2017. The referral decision following PC+, (i.e. referral back to the general practitioner (GP) or referral to outpatient hospital care) was the primary outcome. Stepwise logistic regression modelling was used to describe variations in the referral decisions following PC+, with patient age and gender, number of PC+ consultations, patient diagnosis and treatment specialist as the predicting factors.

Results: A total of 2952 patients visited PC+ for dermatology care. Of those patients with a registered referral, 80.2% (N= 2254) were referred back to the GP, and 19.8% (N= 558) were referred to outpatient hospital care. In the multivariable model, only the treating specialist and patient's diagnosis independently influenced the referral decisions following PC+.

Conclusion: The aim of PC+ is to reduce the number of referrals to outpatient hospital care. According to the results, the treating specialist and patient diagnosis influence referral decisions. Therefore, the results of this study can be used to discuss and improve specialist and patient profiles for PC+ to further optimise the effectiveness of the initiative.

BACKGROUND

Over the course of the last decade, global expenditure on healthcare as a share of world income has been increasing [1-3]. In the coming decades, healthcare spending is even expected to increase faster than prosperity [2, 4]. The population is ageing, and other explanations for rising expenses, such as technological development and lagging productivity, are likely to remain applicable in the future.

As a way to control costs and the utilisation of healthcare services, several countries (e.g. the Netherlands, the UK, Spain and Scandinavian countries) have implemented a gatekeeper system [5-8]. In these systems, general practitioners (GPs) fulfil an important role in patients' further access to healthcare [5]. In addition, hospital care and specialist care (except emergency care) are accessible only upon referral from a GP. Since the literature shows that these systems lead to lower use of health services [9] more appropriate and more effective healthcare use [10] and lower expenditures [11], it is beneficial to further strengthen the position of primary care. Therefore, there have been many attempts to improve the effectiveness and efficiency of primary care and the referral process to outpatient hospital care to strengthen healthcare sustainability [12, 13].

Since 1972, healthcare expenditure as a percentage of the gross domestic product (GDP) has been increasing annually in the Netherlands [14]. Therefore, guaranteeing the financial sustainability of the healthcare system in the future is high on the Dutch political agenda [15].

To provide better care at lower costs, so-called pioneer sites have been appointed by the Minister of Health in the Netherlands [16]. At these pioneer sites, health insurers, care providers and patient organisations join forces to establish initiatives to improve the quality of care and reduce healthcare costs. The main goal of these initiatives is to accomplish the three dimensions of the Triple Aim principle proposed by Berwick et al. [17]. This principle focuses on reducing the per capita cost of healthcare, improving the health of the population and improving the patient experience of care. The 'Blue Care' pioneer site in the Maastricht-Heuvelland region has implemented several initiatives, one of which is Primary Care Plus (PC+). PC+ uses the concept of substitution, which focuses on shifting specialised care to less expensive and more accessible primary care [18]. The aim of PC+ is to achieve substitution by stimulating integrated care through the facilitating of consultations with medical specialists in the primary care setting. Internationally, comparable models of care

are implemented, as for example specialist outreach services and shifted outpatient clinics [12, 13, 19, 20].

One of the specialties involved in PC+ is dermatology. Specialised dermatology care is in high demand due to the increase in the number of patients with dermatological complaints visiting their GPs [21, 22]. Skin conditions are among the most common diseases that are encountered by GPs and for which patients are referred to secondary care [21, 23, 24]. In the Netherlands, 14% of all GP consultations are related to a dermatological disorder [25]. In addition, the number of GP consultations for suspected lesions is increasing by 7.3% annually, and further increases are expected [26, 27]. Along with media campaigns aimed at increasing awareness about the danger of skin cancer and the ageing population [28], the increase in the number of dermatology-related consultations will lead to a growing demand for dermatology-related healthcare services. In addition, GPs often have a lack of dermatological knowledge, which is a reason for diagnostic uncertainty and the experience of difficulties with the diagnosis and treatment of skin disease [29-31]. Moreover, there is large variation in GP referrals to specialised medical care, which is caused by many factors, such as uncertainty about the diagnosis, perceived seriousness of the skin disease and patient preference [32, 33]. GPs' referral decisions are crucial for the patients' progress through the healthcare system and, moreover, for the costs of the healthcare system [34]. Therefore, with PC+, the use of specialist medical expertise in primary care can be strengthened and expanded and unnecessary referrals to (expensive) outpatient hospital care can be avoided.

Because of the novelty of PC+ at its initiation in 2014, clear guidelines for GPs about the exact type of patients and complaints to be referred to PC+ were lacking. Therefore, this study explores referral decisions following PC+ dermatology care and the influence of predictive patient and consultation characteristics. The results of this study could contribute to the development of patient profiles and input for the optimisation of the PC+ process.

METHODS

Design

This retrospective study uses clinical data on referral decisions from patients who received dermatology care in PC+ from January 2015 to March 2017.

Setting

PC+ is an initiative implemented in the pioneer site 'Blue Care', located in the Maastricht-Heuvelland region, in which 81 GPs in 55 GP practices care for a population of approximately 170,000 people [35]. In this region, different organisations work together and developed the PC+ intervention to substitute specialised medical care with primary care [36]. After a pilot, in which medical specialist performed consultations in GP practices, PC+ was implemented on a larger scale with two independent PC+ centres located in the city of Maastricht [37, 38]. This allowed GPs within the region to refer patients to a medical specialist in a neutral primary care setting, with GPs remaining responsible for their patients throughout the whole PC+ care process.

The focus of this study was on dermatology care in the current PC+ setting. Together with orthopaedics, internal medicine, neurology, otolaryngology, ophthalmology, and rheumatology, dermatology has been included in the two PC+ centres from the beginning. Over time, more medical specialties, including paediatrics, gynaecology, urology and a multidisciplinary back pain consultation facility with anaesthesiology and orthopaedics focusing on chronic pain, have been added. Between January 2015 and March 2017, 10,029 patients visited PC+. With 2,952 patients, dermatology accounted for almost one-third of all patients in PC+. The distribution of patients among the different medical specialties is shown in Table 1. The low numbers of patients for some medical specialties were mainly caused by their later influx into PC+ and the lack of personnel for some specialties to organise PC+ consultations on a regular basis.

Table 1 Number of patients visiting Primary Care Plus for the different medical specialties (N=10,029)

Medical specialty	Number of patients % (N)	Start in PC+
Dermatology	29.4% (2,952)	January 2015
Orthopaedics	17.0% (1,708)	January 2015
Internal medicine	2.9% (291)	January 2015
Neurology	6.4% (638)	January 2015
Otolaryngology	18.1% (1,815)	January 2015
Ophthalmology	10.4% (1,044)	January 2015
Rheumatology	5.6% (559)	January 2015
Paediatrics	0.5% (50)	November 2015
Gynaecology	6.6% (659)	December 2015
Urology	1.5% (149)	March 2016
Back pain consultation facility	1.6% (163)	November 2016

Note: PC+ = Primary Care Plus

Intervention

In the PC+ centres, patients with low-complex and non-acute health problems are seen by a medical specialist during a maximum of two consultations, after a referral from their GP. The two PC+ centres operate according to the same method; however, they differ from each other based on the number of consultation hours and the number of different medical specialties. Specialists in PC+ are senior staff specialists working as employees in Maastricht UMC+. The senior staff requirement is part of the specialist profile for PC+, which was established based on previous research [37]. Specialists are paid according to the standard hourly rate. The costs of the space used by the specialist in PC+ is part of the consultation fee. Furthermore, care in PC+ is claimed as primary care performance, through which it can be offered at a lower price compared to secondary care and consultations are not subjected to the patient's deductible.

The process of referring a patient to dermatology care in PC+ is similar to the process of referring a patient to outpatient hospital care and is shown in Figure 1. GPs could refer a patient to PC+ when they had doubts about the diagnosis and/or treatment of

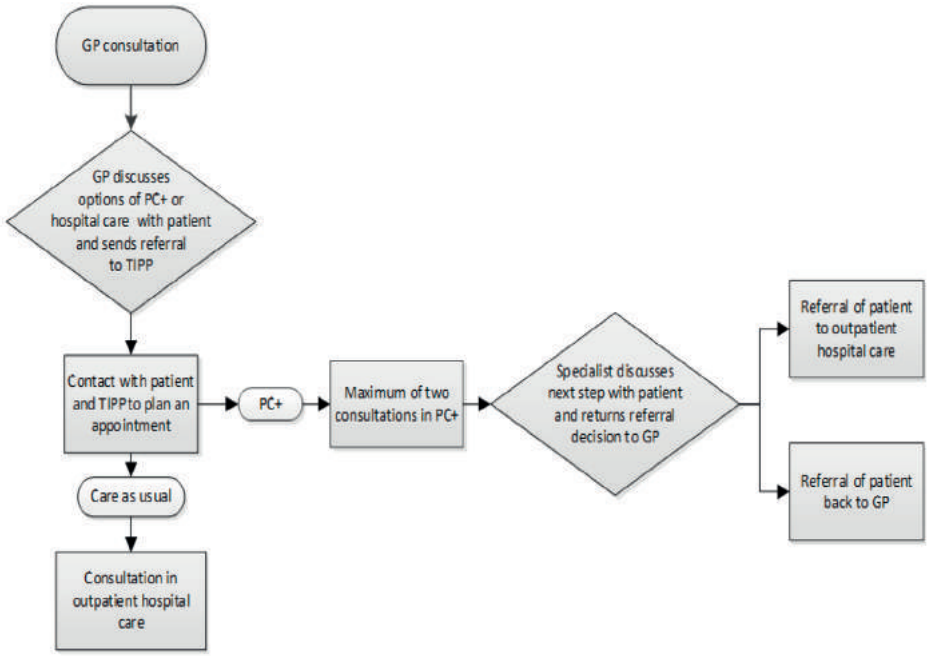

Figure 1 Flow chart of the Primary Care Plus process

patients with, what appeared to be low-complex and non-acute dermatology-related health problems. Profiles for patients eligible for PC+ were formulated by GPs and dermatologists during the study period and were made accessible online for GPs (see Additional file 1). These profiles were based on the experiences of GPs and medical specialists. In addition, it was assumed that patients referred to PC+ would have been referred to outpatient hospital care in a (hypothetical) situation in which PC+ was not available. The final decision to refer a patient to PC+ or to refer to care as usual (outpatient hospital care) was made based on consultation between the GP and the patient. After the decision was made, the referral was first sent to the Transmural Interactive Patient Platform (TIPP), which plans and registers referrals to medical specialists (either in PC+ or outpatient hospital care). In PC+, patients were seen by a dermatologist, and if necessary, dermatologists were able to perform cryotherapy, skin biopsies, blood tests, microbiology and Wood's light investigation. Specialists treated patients and/or provided advice for GPs on further treatment strategies.

In this study, data from all patients visiting PC+ for dermatology care were collected.

Outcome measures

The primary outcome in this study was the referral decision following PC+ (i.e. referral back to the GP or referral to outpatient hospital care). The independent variables were the consultation-related factors: number of PC+ consultations, treating specialist and patient diagnosis. The treating specialist was the specialist who treated the patient during the last PC+ consultation. In addition, the 'treating specialist' variable was divided into four categories: the first three categories included the three specialists who had performed the most PC+ consultations, and the fourth category included all other dermatologists working in PC+. The three specialists who had performed the most PC+ consultations had worked in PC+ since the beginning of the study period (January or February 2015). The specialists in the 'other dermatologists' category had started working in PC+ at some point during the study period (between January 2015 and October 2016). Patient diagnosis was defined as the diagnosis determined by the specialist during the last PC+ consultation according to the International Classification of Diseases (ICD-10) [39]. This variable was divided into mutually exclusive categories (meaning that patients could be placed in only one diagnosis category): the first ten categories included the ten most common diagnoses in PC+, an 11th category included all other diagnoses and a 12th category was for unknown diagnosis. The corresponding ICD-10 codes of the ten most common diagnoses in PC+ are presented in Additional file 2. In addition, patient age (in years) and gender were used.

Statistical analysis

Continuous data are presented as the means and standard deviations (SDs). Categorical data are presented as the counts and percentages. Consultation-related factors and patient-related factors were compared between the two possible referral decisions following a PC+ consultation: (1) referral back to the GP or (2) referral to outpatient hospital care. An independent-samples t-test was used to compare the continuous data, and Pearson's x^2 test was used to compare the categorical data. P-values ≤ 0.05 were considered statistically significant.

To describe variations in referral decisions, stepwise logistic regression modelling was used, with the decision to refer to outpatient hospital care as a binary yes/no variable. First, univariate logistic regression analysis was used to evaluate the relation between the primary outcome and the independent variables (predictors). Predictors with a p-value of ≤0.15 were included in the multivariable logistic regression analysis. For categorical variables, the variable was included when one or more categories had a p-value of ≤0.15. In this multivariable model, backwards

elimination of the included variables was performed. The results were presented as unadjusted and adjusted odds ratios (ORs and AORs, respectively) with 95% confidence intervals (95% CIs), supplemented by the average marginal effects (AMEs). AMEs represent the difference in the adjusted predictions of the dependent variable relative to the reference group and improve the interpretability of the results [40]. With regard to the categorical variables treating specialist and diagnosis, the category within these variables that had an outpatient hospital care referral rate that was closest to the total average of that variable and that had a reasonable sample size was selected as the reference group. The explained variation in the regression model was measured by the Nagelkerke pseudo R^2 [41].

Analyses were performed using SPSS software for Windows version 24.0 (SPSS Inc., Chicago, IL, USA) and R Studio (R Studio, Boston, MA).

RESULTS

Between January 2015 and March 2017, 2,952 patients visited PC+ for dermatology care. The referral decision following PC+ was unknown for 140 patients; therefore, these patients were excluded from the analysis. These patients did not differ from the included patients in terms of age or gender ($p = 0.748$ and $p = 0.430$, respectively) (see Additional file 3). However, the excluded patients had significantly fewer PC+ consultations ($p = 0.009$). Furthermore, there was a difference in the distribution of treating specialists and diagnoses between the included and excluded patients ($p = 0.002$ and $p \leq 0.001$, respectively).

The remaining 2,812 patients had a total of 3,355 PC+ consultations (average of 1.19, SD = 0.4 consultations). Following PC+, 80.2% (N = 2,254) of the patients were referred back to their GPs, and 19.8% (N = 558) were referred to outpatient hospital care for further treatment/examination (see Table 2).

PC+ patients referred to outpatient hospital care were significantly older than those referred back to their GPs ($p \leq 0.001$). There was no significant difference between the two groups with regard to gender ($p = 0.563$). PC+ patients referred to outpatient hospital care had significantly fewer PC+ consultations ($p = 0.045$). However, the difference was very small and therefore was not very clinically relevant. In addition, there were differences in the distribution of the referral decision by treating specialist and diagnosis within PC+ (both $p \leq 0.001$).

Table 2 Overview and comparisons of Primary Care Plus patients and consultation characteristics

	Tota (N = 2,812)	**Referred back to GP 80.2% (N = 2,254)**
Age in years (mean ± SD)	47.7 ± 20.9	46.8 ± 21.0
Gender – male % (N)	41.2 (1,159)	40.9 (923)
Number of consultations (mean ± SD)	1.19 ± 0.4	1.20 ± 0.4
Specialist		
Specialist 1 % (N)	53.6 (1,508)	84.6 (1,276)
Specialist 2 % (N)	25.6 (721)	76.0 (548)
Specialist 3 % (N)	11.1 (311)	72.3 (225)
Other % (N)	9.7 (272)	75.4 (205)
Diagnosis		
Naevi % (N)	14.5 (407)	77.9 (317)
Premalignant dermatosis %(N)	9.4 (264)	81.8 (216)
Benign tumours % (N)	8.5 (238)	84.9 (202)
Other eczema% (N)	7.8 (219)	93.2 (204)
Acneiform dermatoses % (N)	6.1 (172)	87.2 (150)
Inflammatory dermatoses % (N)	5.7 (161)	70.2 (113)
Dermatoses due to microorganisms % (N)	5.3 (149)	93.3 (139)
Malignant dermatoses % (N)	5.2 (146)	26.0 (38)
Hair and nail disorders % (N)	3.7 (103)	95.1 (98)
Pigment disorders % (N)	3.3 (94)	85.1 (80)
Other % (N)	23.2 (653)	85.9 (561)
Unknown % (N)	7.3 (206)	66.0 (136)

Note: GP = general practitioner; SD = standard deviation

** P < 0.05; ** P < 0.001*

Specialists

In total, 12 different dermatologists worked in PC+ during the study period. However, the number of PC+ consultations held by these specialists varied greatly. There were three specialists who had seen the vast majority of patients. These three specialists saw approximately 90% (N = 2,540) of the patients visiting PC+ for dermatology care during the study period.

Predictors of a referral to outpatient hospital care

The results of the univariate and multivariable logistic regression analyses are shown

	Referred to hospital care 19.8% (N = 558)	p-values
	51.5 ± 20.4	≤0.001**
	42.3 (236)	0.563
	1.16 ± 0.4	0.045*
		≤0.001**
	15.4 (232)	
	24.0 (173)	
	27.7 (86)	
	24.6 (67)	
		≤ 0.001**
	22.1 (90)	
	18.2 (48)	
	15.1 (36)	
	6.8 (15)	
	12.8 (22)	
	29.8 (48)	
	6.7 (10)	
	74.0 (108)	
	4.9 (5)	
	14.9 (14)	
	14.1 (92)	
	34.0 (70)	

in Table 3. Variables with a p-value ≤ 0.15 in the univariate analysis (age, number of consultations, treating specialist and diagnosis) were included in the multivariable logistic regression analysis. In the multivariable model only, treating specialist and patient diagnosis were retained as variable that independently influenced a referral to outpatient hospital care.

Patients treated by specialist 2 (AOR 1.88, 95% CI = 1.48–2.39, AME = 0.09), specialist 3 (AOR 1.97, 95% CI = 1.44–2.69, AME = 0.09) or another (less common)

specialist (AOR 1.80, 95% CI = 1.29–2.52, AME = 0.08) were more likely to be referred to outpatient hospital care following PC+ than patients treated by specialist 1 (reference group). In addition, patients diagnosed with malignant dermatosis (AOR 12.98, 95% CI = 7.96-21.17, AME = 0.55) or inflammatory dermatoses (AOR 2.12, 95% CI = 1.33-3.38, AME = 0.14) and patients for whom the diagnosis was unknown (AOR 2.24, 95% CI = 1.45-3.45, AME = 0.15) were more likely to be referred to outpatient hospital care than patients diagnosed with premalignant dermatosis (reference

Table 3 Logistic regression analysis of referral to outpatient hospital care among dermatology patients in Primary Care Plus (N = 2,812)

Predictors	Univariable model		
	Unadjusted OR (95% CI)	AME	*p*-value
Age [A]	1.12 (1.07-1.17)	0.02	≤0.001***
Gender - male	1.06 (0.88-1.28)	0.01	0.510
Number of consultations	0.79 (0.62-1.00)	-0.04	0.054
Specialist			
Specialist 1	0.55 (0.45-0.66)	-0.10	≤0.001***
Specialist 2	1.40 (1.14-1.72)	0.06	≤0.001***
Specialist 3	1.64 (1.26-2.15)	0.09	≤0.001***
Other specialists	1.36 (1.02-1.83)	0.05	0.038*
Diagnosis			
Naevi	1.18 (0.91-1.52)	0.03	0.215
Premalignant dermatosis	0.89 (0.64-1.23)	-0.02	0.477
Benign tumours	0.70 (0.49-1.01)	-0.05	0.058*
Other eczema	0.28 (0.16-0.47)	-0.14	≤0.001***
Acneiform dermatoses	0.58 (0.36-0.91)	-0.08	0.018*
Inflammatory dermatoses	1.78 (1.26-2.53)	0.11	≤0.001***
Dermatoses due to microorganisms	0.28 (0.15-0.53)	-0.14	≤0.001***
Malignant dermatoses	14.00 (9.54-20.53)	0.57	≤0.001***
Pigment disorders	0.70 (0.39-1.24)	-0.05	0.223
Hair and nail disorders	0.20 (0.09-0.49)	-0.16	≤0.001***
Other diagnosis	0.60 (0.47-0.76)	-0.07	≤0.001***
Unknown diagnosis	2.23 (1.65-3.03)	0.15	≤0.001***

Note: OR = odds ratio; CI = confidence interval; AME = average marginal effects

** P ≤ 0.15; ** P < 0.01; *** P < 0.001; [A] Age was rescaled such that one unit is equal to 10 years; [B] Variable not significant in final model; [C] Reference category for the adjusted OR analysis*

group). On the other hand, patients diagnosed with other eczema (AOR 0.36, 95% CI = 0.19-0.66, AME = -0.11), dermatoses due to microorganisms (AOR 0.32, 95% CI = 0.16-0.66, AME = -0.11) and hair and nail disorders (AOR 0.23, 95% CI = 0.09-0.59, AME = -0.13) were less likely to be referred to outpatient hospital care following PC+ consultations. The final model explained 19.3% of the variation in PC+ referral decisions (Nagelkerke R^2 = 0.193).

	Final model		
	Adjusted OR (95% CI)	**AME**	***p*-value**
	…[B]		
	…		
	…[B]		
	…[C]		
	1.88 (1.48-2.39)	0.09	≤0.001***
	1.97 (1.44-2.69)	0.09	≤0.001***
	1.80 (1.29-2.52)	0.08	≤0.001***
	1.30 (0.88-1.92)	0.04	0.195
	…[C]		
	0.83 (0.51-1.33)	-0.03	0.433
	0.36 (0.19-0.66)	-0.11	≤0.001***
	0.66 (0.38-1.14)	-0.05	0.136
	2.12 (1.33-3.38)	0.14	0.002**
	0.32 (0.16-0.66)	-0.11	0.002**
	12.98 (7.96-21.17)	0.55	≤0.001***
	0.77 (0.40-1.49)	-0.03	0.441
	0.23 (0.09-0.59)	-0.13	0.002**
	0.71 (0.49-1.05)	-0.04	0.086
	2.24 (1.45-3.45)	0.15	≤0.001***

DISCUSSION

This study explored referral decisions following dermatology care in PC+ and the influence of predictive patient and consultation characteristics on this decision. The results showed that the majority of the patients (80.2%) were referred back to their GPs following a consultation for dermatology care in PC+. This finding is in line with previous research suggesting that initiatives like PC+ have the ability to reduce outpatient hospital care referrals and/or increase the appropriateness of referrals made [13]. However, it is important to verify whether the assumption based on previous research that all patients would have been referred to secondary care if PC+ had not been available is also valid in this case [38].

Furthermore, the results showed that the treating specialist and patient diagnosis independently influenced referral decisions following dermatology care in PC+. Regarding the treating specialist, previous research by van Hoof et al. [37] indicated a profile for appropriate specialists in PC+. According to this profile, specialists should, in addition to having a certain degree of seniority, work according to a generalist approach and have an attitude that is consistent with the model of substitution. The extent to which the included specialists met this profile was not part of this study. However, the results indicated that the likelihood of patients being referred to outpatient hospital care was influenced by the treating specialists. A reason for this could be that the ability to work in a PC+ setting differs among specialists, and for example, specialists with a less generalist approach may refer patients to outpatient hospital care more often. Therefore, more research is needed to study the ability of specialists to work in PC+.

Regarding the patient diagnosis, the results provide an indication of diagnoses that are suitable for PC+. However, high referral rates to outpatient hospital care do not necessarily indicate that complaints leading to these diagnoses are inappropriate for PC+. For example, regarding malignant dermatoses, PC+ can function as a screening tool to prevent patients with an unjustified suspicion from being referred to hospital care for unnecessary testing and treatment. In addition, PC+ can improve early detection, and patients with more suspicious symptoms can be referred to secondary care for treatment, which may reduce mortality and improve quality of life [42]. In addition, diagnoses such as other eczema, dermatoses due to microorganisms and hair and nail disorders, which have low referral rates to outpatient hospital care, seem particularly suitable for PC+. Nevertheless, these diagnoses will not necessarily always be appropriate for PC+. GPs may also experience a (too) low threshold when referring patients to PC+ [37].

As suggested by van Hoof et al. [37], GPs and specialists should discuss appropriate and inappropriate complaints, symptoms and diseases for PC+. The results of this study can provide input for this discussion and can be used to further develop patient profiles for PC+ (see Additional file 1) if necessary. In addition, when a patient profile for PC+ is composed, appropriate and inappropriate diagnoses should be translated into the International Classification of Primary Care (ICPC) codes [43]. The ICPC-codes are used by every Dutch GP and function to categorise patients' complaints, symptoms and diseases. One specific ICPC-code could ultimately lead to several diagnoses. In this study, the ICPC-codes were not available. Therefore, the patient diagnosis made by the specialist in PC+ was used as a predictor of the referral decision. Furthermore, the clear provision of advice from specialists to GPs when specialists refer patients back to GPs could contribute to a learning effect among GPs regarding the diagnosis or treatment of dermatology patients and whether to refer to PC+ or outpatient hospital care [44]. This feedback could contribute to bridging the knowledge gap between primary and secondary care [29].

The variation explained by the final model in this study was 19.3%, which implies that a lot of variation is explained by other (party unknown) variables that were not included in the model. According to the literature on GP referrals to hospital care, case-specific factors, such as the nature of the disease and the observed severity, influence the patient referrals [45]. In addition, other patient-associated factors than age and sex, such as the overall health status, insurance coverage, social class, needs and values, pressure for referral and preferences, may influence the referral decision [33, 32, 46].

The results showed that 29.4% of all patients visiting PC+ during the study period had dermatological complaints. This percentage is higher than the 14% of all Dutch patients visiting their GP with a dermatological complaint. However, it is difficult to compare these percentages, since not all medical specialties are represented in PC+. In addition, the medical specialties in PC+ did not have an equal number of consultation hours during the study period due to an unequal influx of patients and a lack of personnel for some medical specialties. Finally, PC+ is focused on a select group of patients with low-complex and non-acute dermatology related health problems who are eligible for this care (see Additional file 1).

Moreover, since PC+ focuses on non-acute and low-complex care, it is assumed that more serious diagnoses, such as malignancies, are not made more often

in PC+ than in outpatient hospital care. Epidemiological data on dermatological conditions in primary care and hospital care in the Netherlands [47] and the data for patients diagnosed with malignant dermatoses in the present study are consistent with this observation. In PC+, 5.2% of the patients were diagnosed with malignant dermatoses, compared to 12% of patients in hospital care [47] (see Additional file 4). On the other hand, it is assumed that less serious diagnoses are made more often in PC+ than in outpatient hospital care, since the aim of PC+ is to substitute secondary care with primary care for low-complex care. Based on epidemiological data and the data in the present study, it can be concluded that this is the case for diagnoses such as naevi, benign tumours and inflammatory dermatoses.

In addition, there are various other approaches to reduce outpatient hospital care referrals and/or increase the appropriateness of referrals [13], such as the concept of teledermatology [22, 48] and the employment of GPs with special interests and the implementation of nurse-led services in these kind of settings [49, 50, 23]. Even though these initiatives show generally positive findings in terms of accessibility, waiting time and patient satisfaction [51, 52, 22], researchers also have critiqued the diagnostic accuracy of telemedicine [53], the lack of specific research on patient safety [54], and the limited evidence regarding cost-effectiveness [55].

LIMITATIONS

The use of monitoring data limited the amount of information, and therefore predictors, for this study. Extending the data, for example with data from GP practices, can generate more useful information. Examples include the ICPC codes and registration of the severity of the complaints. By expanding the data, the referral decision may be better predicted and more and better information can be given back to GPs and medical specialists in order to improve the efficiency of PC+. In this study, data expansion was not possible because data from GP practices in this region are registered through different systems, which makes data linking complicated.

Additionally, a limited number of specialists were included in the present study. However, differences in the referral decisions of these specialists were observed. It was not possible to include more characteristics of the PC+ specialists in the present study since these variables may affect the anonymity of the specialists involved. However, the results of this study can be used as input for further research. Including more specialists and more characteristics in further research, such as specialist

age and work experience, could contribute to more insight into the variation in specialists' referrals and, therefore, to more input for PC+ the specialist profiles.

Furthermore, follow-up data of patients visiting PC+ for dermatology care were not taken into account. It is possible that patients who were initially referred back to their GPs had follow-up visits for the initial complaint in secondary care shortly having a PC+ consultation. If this pattern were to occur on a significant scale, PC+ would be less appropriate. Therefore, hospital data should be analysed. It is also relevant to determine whether the substitution effect is present in outpatient hospital care.

Finally, the referral decisions following the PC+ consultations of 140 patients and the diagnoses of 206 patients were missing. The results showed that patients excluded from the analysis differed from the included patients in terms of the number of PC+ consultations, and the distribution of treating specialists and diagnoses; therefore, selection bias may exist (see Additional file 3). However, only 140 of the 2,952 patients needed to be excluded, which is a relatively low number. Furthermore, incomplete patient cases were partly caused by specialists becoming accustomed to the registration method at the beginning of PC+. Therefore, the degree of selection bias seems limited and it is not expected that the results were considerably influenced.

CONCLUSION

To conclude, through the referral of a large number of patients back to their GPs following dermatology care in PC+, the number of referrals to hospital care can be limited; thus, dermatology care seems to be suitable for PC+. Both the treating specialist and the patient diagnosis influenced the referral decision. Therefore, the results of this study can be used to discuss and improve profiles for specialists and patients in PC+ to further optimise the effectiveness of the initiative. Besides insight into the influence on quality of care, further research is needed into the costs and volumes of dermatology care, both in PC+ and secondary care to determine if substitution of dermatology care actually occurs and healthcare costs are reduced.

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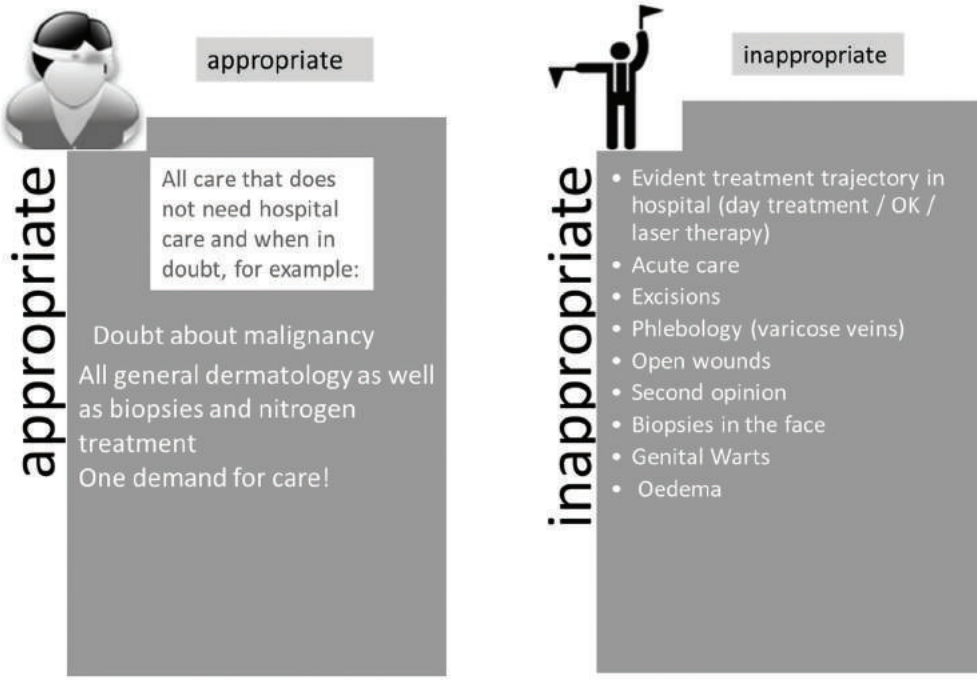
ADDITIONAL FILES

Additional file 1 Dermatology patient profile of Primary Care Plus



Additional file 2 Additional file 2 Diagnosis and corresponding ICD-10

Table 1 Top ten diagnoses in Primary Care Plus with corresponding International Classification of Diseases (ICD-10) codes

Naevi				
D22.3	D22.90	D22.916	D22.96	D23.9
D22.312	D22.91	D22.92	D22.97	D48.5
D22.512	D22.911	D22.93	D22.971	L81.41
D22.513	D22.914	D22.94	D22.98	L81.410
D22.9	D22.915	D22.95	D22.99	
Premaligne dermatosis				
A63.8	D03.9	D07.4	D47.0	L44.82
C90.2	D04.9	D29.9	D48.1	L57.0
Benign tumours				
D17.9	D21.91	D23.33	D23.914	D23.92
D18.1	D21.92	D23.90	D23.915	D23.920
D21.14	D21.93	D23.91	D23.916	D23.921
D21.5	D23.0	D23.911	D23.917	D23.922
D21.9	D23.24	D23.913	D23.918	D23.923
D21.91	D23.90	D23.916	D23.922	D23.93
Other eczema				
I83.1	L30.11	L30.4	L30.82	L30.86
L30.0	L30.12	L30.5	L30.84	L30.87
L30.1	L30.13	L30.8	L30.85	L30.88
L30.11				
Acneiform dermatoses				
L70.0	L70.05	L70.4	L70.81	L70.85
L70.01	L70.1	L70.41	L70.82	L70.86
L70.02	L70.2	L70.5	L70.83	L70.9
L70.03	L70.3	L70.8	L70.84	L71.0
L70.04				
Inflammatory dermatoses				
D69.0	D89.1	L10.8	L13.9	L43.3
D69.01	H61.0	L12.0	L21.11	L43.8
D69.02	I89.8	L12.1	L22.0	L43.81
D69.8	K12.0	L12.11	L22.1	L43.9
D86.3	L10.0	L12.2	L22.2	L44.1
D86.31	L10.1	L12.21	L30.42	L44.2

L81.45	Q82.2	Q82.53	Q82.57	Q82.810
L81.46	Q82.21	Q82.54	Q82.58	Q82.811
L81.47	Q82.222	Q82.55	Q82.59	Q82.813
L81.48	Q82.3	Q82.56	Q82.591	Q82.9

L57.1	L90.0	L90.02	N89.4	N90.4
L85.85	L90.01	N48.0		

D23.924	D23.930	D23.98	D76.3	L72.1
D23.927	D23.931	D23.99	D76.31	L72.2
D23.928	D23.95	D24.0	K13.71	L72.8
D23.929	D23.96	D36.1	L72.0	L72.81
D23.93	D23.97	D36.11	L72.01	L72.9

L30.91	L56.1	L56.8	L56.9	L56.92
L55.9	L56.4	L56.81	L56.91	L85.31
L56.0	L56.41	L56.82		

L71.01	L71.81	L71.9	L73.1	L73.82
L71.02	L71.82	L71.91	L73.2	L73.83
L71.1	L71.83	L72.82	L73.8	L73.84
L71.8	L71.84	L73.0	L73.81	L73.9

L52.1	L95.1	M08.2	M32.9	M35.2
L52.2	L95.8	M15.1	M33.0	M35.3
L52.3	L95.9	M30.0	M33.1	M35.4
L57.01	L98.2	M30.3	M34.1	N48.1
L57.5	L98.3	M31.3	M34.8	N48.11
L92.0	L98.8	M31.4	M34.81	N48.12

Table 1 Continued

Inflammatory dermatoses				
D86.32	L10.2	L12.3	L40.11	L51.0
D86.33	L10.21	L13.0	L43.0	L51.1
D86.34	L10.4	L13.1	L43.1	L51.2
D86.8				
Dermatoses due to microorganism				
A01.0	A30.5	A59.01	A75.9	B08.1
A06.7	A30.8	A59.02	A77.0	B08.2
A18.40	A30.81	A59.8	A77.1	B08.3
A18.41	A30.82	A59.9	A77.9	B08.4
A18.42	A30.9	A65.	A79.0	B08.5
A18.43	A31.1	A65.1	A79.1	B08.8
A18.44	A31.11	A66.0	A79.9	B08.81
A18.45	A31.12	A66.1	A90.0	B09.0
A18.46	A31.9	A66.3	B00.0	B16.9
A18.47	A35.0	A66.4	B00.1	B26.9
A18.48	A36.3	A66.5	B00.2	B27.9
A18.49	A38.0	A66.6	B00.8	B34.9
A20.0	A39.1	A66.7	B00.9	B35.0
A20.1	A39.4	A66.8	B01.9	B35.1
A22.0	A40.9	A66.9	B02.2	B35.11
A24.0	A41.0	A67.9	B02.3	B35.2
A26.0	A41.8	A69.1	B02.7	B35.3
A28.1	A41.9	A69.2	B02.9	B35.4
A30.0	A42.8	A69.21	B03.0	B35.6
A30.1	A46.0	A75.0	B05.9	B35.8
A30.2	A48.0	A75.1	B06.9	B35.81
A30.3	A48.8	A75.2	B08.0	B35.9
A30.4	A59.0	A75.3	B08.01	B36.0
Malignant dermatoses				
B21.0	C44.9	C44.93	C44.98	C49.92
C21.0	C44.90	C44.94	C44.99	C49.93
C21.8	C44.91	C44.95	C46.0	C49.94
C43.6	C44.911	C44.96	C49.9	C49.95
C43.7	C44.92	C44.97	C49.91	C49.96
C43.9				

L93.0	M05.0	M31.6	M34.9	N48.2
L93.1	M05.2	M31.8	M35.0	O26.4
L93.2	M06.3	M32.0	M35.1	P00.8

B36.1	B42.9	B85.1	L02.91	L98.81
B36.2	B43.0	B85.2	L02.92	L98.82
B36.3	B43.2	B85.3	L02.93	N45.9
B36.8	B45.2	B86.0	L03.0	N72.1
B37.0	B47.0	B86.1	L03.01	N75.1
B37.2	B48.0	B87.0	L03.03	N76.0
B37.21	B48.1	B88.0	L03.04	N76.01
B37.22	B55.0	B88.01	L03.9	N76.2
B37.23	B55.1	B88.1	L03.94	N76.3
B37.24	B55.2	B88.8	L04.9	N76.41
B37.25	B55.9	B88.9	L05.0	N89.8
B37.3	B58.9	B96.5	L05.9	P37.5
B37.4	B65.3	H00.0	L08.0	P38.0
B37.8	B73.0	H00.1	L08.02	T81.4
B37.81	B74.0	H00.11	L08.03	T81.41
B37.82	B74.1	I74.9	L08.04	T81.42
B37.83	B74.3	J02.9	L08.1	T88.1
B37.84	B74.8	L00.0	L08.8	
B37.9	B74.9	L00.1	L08.92	
B38.3	B76.9	L01.0	L30.9	
B39.9	B78.1	L01.02	L44.4	
B40.9	B80.0	L01.1	L44.8	
B41.9	B85.0	L02.9	L88.1	

C49.97	C79.8	C84.41	C84.52	C95.9
C50.0	C81.9	C84.5	C90.0	D47.7
C69.9	C84.0	C84.51	C91.9	D04
C77.9	C84.1	C85.1	C92.9	D72.8
C79.2	C84.4	C85.9	C93.9	D76.0

Table 1 Continued

Hair and nail disorders				
E70.32	L60.1	L60.36	L60.84	L65.0
E70.33	L60.2	L60.37	L60.85	L65.01
H30.8	L60.3	L60.38	L60.86	L65.1
L01.01	L60.30	L60.4	L60.9	L65.11
L01.03	L60.31	L60.5	L63.0	L65.2
L08.81	L60.32	L60.8	L63.2	L65.8
L21.02	L60.33	L60.81	L63.9	L65.81
L58.11	L60.34	L60.82	L64.9	L65.82
L60.0	L60.35	L60.83		
Pigment disorders				
E70.3	L81.01	L81.21	L81.411	L81.44
E70.31	L81.1	L81.3	L81.42	L81.49
L81.0	L81.2	L81.4	L81.43	L81.5

L65.83	L66.81	L67.83	Q84.11	Q84.23
L65.9	L66.9	L67.9	Q84.12	Q84.3
L65.91	L67.0	L67.91	Q84.13	Q84.31
L65.92	L67.1	L68.0	Q84.14	Q84.4
L66.1	L67.11	L68.1	Q84.15	Q84.5
L66.2	L67.8	L68.9	Q84.16	Q84.6
L66.3	L67.81	Q84.0	Q84.2	Q84.61
L66.8	L67.82	Q84.1	Q84.22	Q84.62

L81.6	L81.62	L81.7	L81.72	L81.81
L81.61	L81.63	L81.71	L81.8	L81.9

Additional file 3 Comparison of patient categories

Table 1 Comparison of patients and consultation characteristics of included and excluded patients

	Included patients (N = 2,812)	**Excluded patients (N = 140)**	**p-values**
Age in years (mean ± SD)	47.7 ± 20.9	48.3 ± 20.6	0.748
Gender – male % (N)	41.2 (1,159)	37.9 (53)	0.430
Number of consultations (mean ± SD)	1.19 ± 0.4	1.10 ± 0.40	0.009*
Specialist			0.002*
Specialist 1 % (N)	53.6 (1,508)	48.6 (68)	
Specialist 2 % (N)	25.6 (721)	27.1 (38)	
Specialist 3 % (N)	11.1 (311)	5.7 (8)	
Other % (N)	9.7 (272)	18.6 (26)	
Diagnosis			≤0.001**
Naevi % (N)	14.5 (407)	2.1 (3)	
Premalignant dermatosis % (N)	9.4 (264)	15.0 (21)	
Benign tumours % (N)	8.5 (238)	2.1 (3)	
Other eczema % (N)	7.8 (219)	14.3 (20)	
Acneiform dermatoses % (N)	6.1 (172)	9.3 (13)	
Inflammatory dermatoses % (N)	5.7 (161)	10.0 (14)	
Dermatoses due to microorganism % (N)	5.3 (149)	6.4 (9)	
Malignant dermatoses % (N)	5.2 (146)	1.4 (2)	
Hair and nail disorders % (N)	3.7 (103)	0.7 (1)	
Pigment disorders % (N)	3.3 (94)	7.9 (11)	
Other % (N)	23.2 (653)	26.4 (37)	
Unknown % (N)	7.3 (206)	4.3 (6)	

Note: SD = standard deviation

** P < 0.01; ** P ≤ 0.001*

Additional file 4 Distribution of dermatological complaints

Table 1 Comparison of the distribution of dermatological complaints in primary care, Primary Care Plus and secondary care

Diagnosis	Primary care (General Practitioner) %	Primary Care Plus %	Secondary care (outpatient hospital care) %
Naevi	3.0	14.5	9.0
Premaligne dermatosis	-	9.4	10.0
Benign tumours	9.0	8.5	7.0
Other eczema*	15.0	7.8	12.0
Acneiform dermatoses	2.0	6.1	6.0
Inflammatory dermatoses	18.0	5.7	4.0
Dermatoses due to microorganisms	-	5.3	-
Malignant dermatoses	1.0	5.2	12.0
Hair and nail disorders	-	3.7	-
Pigment disorders	-	3.3	-
Other	52.0	23.2	40.0

** In primary care and secondary care, the category eczema was not further specified.*

03

Referral decisions and its predictors related to orthopaedic care

A retrospective study in a novel primary care setting

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ABSTRACT

Due to the ageing population, the prevalence of musculoskeletal disorders will continue to rise, as well as healthcare expenditure. To overcome these increasing expenditures, integration of orthopaedic care should be stimulated. The Primary Care Plus (PC+) intervention aimed to achieve this by facilitating collaboration between primary care and the hospital, in which specialised medical care is shifted to a primary care setting. The present study aims to evaluate the referral decision following orthopaedic care in PC+ and in particular to evaluate the influence of diagnostic tests on this decision. Therefore, retrospective monitoring data of patients visiting PC+ for orthopaedic care was used. Data were divided into two periods; P1 and P2. During P2, specialists in PC+ were able to request additional diagnostic tests (such as ultrasounds and MRIs). A total of 2,438 patients visiting PC+ for orthopaedic care were included in the analysis. The primary outcome was the referral decision following PC+ (back to the general practitioner (GP) or referral to outpatient hospital care). Independent variables were consultation- and patient-related predictors. To describe variations in the referral decision, logistic regression modelling was used. Results show that during P2, significantly more patients were referred back to their GP. Moreover, the multivariable analysis show a significant effect of patient age on the referral decision (OR 0.86, 95% CI = 0.81–0.91) and a significant interaction was found between the treating specialist and the period ($p = 0.015$) and between patient's diagnosis and the period ($p \leq 0.001$). Despite the significant impact of the possibility of requesting additional diagnostic tests in PC+, it is important to discuss the extent to which the availability of diagnostic tests fits within the vision of PC+. In addition, selecting appropriate profiles for specialists and patients for PC+ are necessary to further optimise the effectiveness and cost of care.

INTRODUCTION

Problems related to the musculoskeletal system are the second most common causes of disability and affect more than 1.7 billion people worldwide [1]. Most musculoskeletal disorders are associated with severe long-term pain and physical disability that affects an individual's daily life [2-4] and are a major cause of work disability and absence, which leads to loss of productivity [2, 3, 5, 6]. Due to the ageing population in developed countries, the prevalence of musculoskeletal disorders will rise [7]. This increasing prevalence will lead to higher demand for health care services since individuals with musculoskeletal problems are among the highest users of care [8] and to a rise in healthcare costs [9].

In the Netherlands, musculoskeletal problems are a significant factor in high healthcare costs [10], with 1.1 billion euros spent on osteoarthritis in 2011, corresponding to 1.2% of total healthcare costs [11]. Most general practice consultations are also related to problems of the musculoskeletal system [12, 13]. Because Dutch general practitioners (GPs) have a gatekeeping role [14], one of the challenges facing them is deciding when to refer patients with musculoskeletal problems and to which medical speciality [15]. A study by Roland et al. [16] showed that, according to medical specialists, almost 25% of GP referrals to orthopaedics were unnecessary and primary care management was more appropriate. GP competency also varies significantly with respect to diagnosing and treating musculoskeletal disorders [17]. Due to rapid developments in diagnostics and treatment, it is unrealistic for GPs to be constantly informed about all possibilities [18]. These knowledge gaps in primary care may lead to referrals to outpatient hospital care for diagnosis and/or treatment that GPs could actually provide if they had the right resources, training, and support [19].

To overcome this gap and to keep patients out of the hospital, the Dutch pioneer site 'Blue Care' implemented Primary Care Plus (PC+) to share and embed specialist knowledge into primary care [20, 21]. PC+ involves hospital specialists providing consultations in the primary care setting, with a minimum of diagnostic tools, to prevent unnecessary referrals to outpatient hospital care. With this, outpatient hospital care is shifted to a more accessible primary care setting [22, 23].

Considering the novelty of PC+, this study examines whether orthopaedic care is suitable to be shifted to the primary care setting. Therefore, this study aims to evaluate the referral decision following orthopaedic care in PC+ in order to determine

to what extent patients are referred back to their GP or how often a referral to hospital care is still necessary. In particular, this study will focus on the influence of the availability of diagnostic tests in PC+ on this referral decision, since contradictory results have been found in literature when it comes to the effect of a lack of diagnostic tools in orthopaedic care [21, 24]. Furthermore, other predictors, like consultation- and patient-related predictors of this decision will be studied as well. With these insights, PC+ for orthopaedic care can be further optimised.

MATERIALS AND METHODS

Design

The present retrospective study makes use of data on referral decisions during the period January 2015 to December 2017. The data were divided into two periods, P1 (from January 2015 to December 2016) and P2 (from January 2017 to December 2017). This distinction was based on the introduction of the possibility of orthopaedic surgeons, working in PC+, requesting additional diagnostic tests (such as ultrasounds and MRIs).

Setting

In pioneer site 'Blue Care' in the Maastricht-Heuvelland region, one of nine pioneer sites in the Netherlands, the primary care organisation Care in Development (in Dutch '*Zorg in Ontwikkeling*'), the Maastricht University Medical Centre+ (Maastricht UMC+), the health insurance company VGZ, and the patient representative foundation House of Care (in Dutch '*Huis voor de Zorg*') work together. The Maastricht-Heuvelland region consists of 81 GPs working in 55 different GP practices caring for a population of about 170,000 people [25].

The Dutch healthcare system is characterised by the gatekeeping principle, meaning that a referral from the GP is required for hospital and specialist care, with the exception of emergency care [26]. Primary care, including GP consultation, is freely accessible for patients [27].

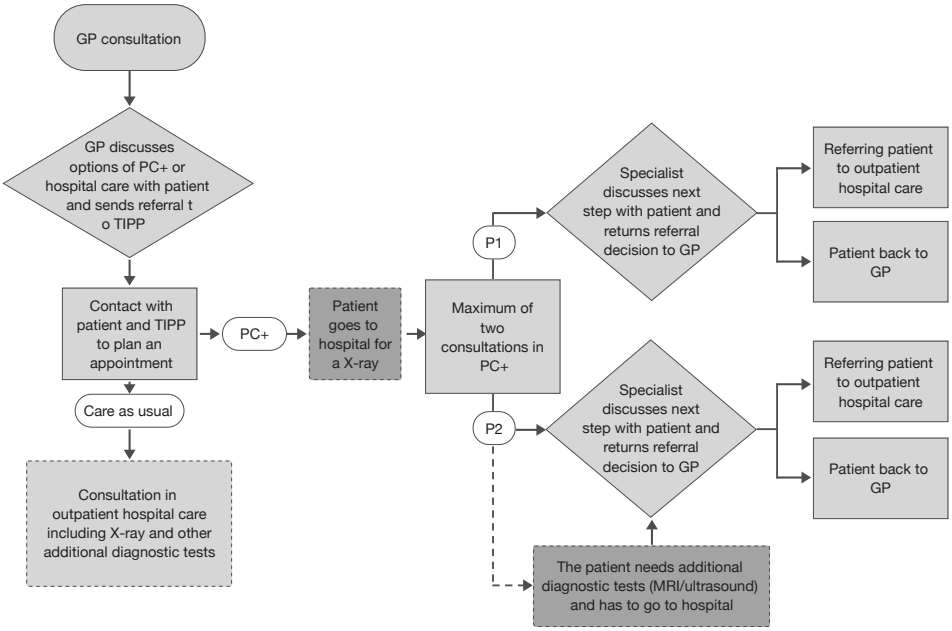
The region Maastricht-Heuvelland developed the PC+ intervention to substitute primary care for outpatient hospital care. The concept of PC+ started with a pilot in which four medical specialties performed consultations within GP practices [21, 28]. Orthopaedic care was one of the four specialties involved. Although the results of the feasibility study by van Hoof et al. [28] showed that PC+ seemed to be

a promising intervention, problems of inefficiency and competitive restraints were found. Therefore, two independent PC+ centres located in the city of Maastricht were established in 2014. These two centres are located outside the hospital site, and the PC+ concept is quite similar to the well-known concept of specialist outreach clinics [29]. With the arrival of the PC+ centres, GPs within the region were able to refer patients to a medical specialist in a neutral primary care setting. The focus of this study was on orthopaedic care in the current PC+ setting.

Intervention

Figure 1 shows the total PC+ process, starting from referral to PC+ to the referral decision made by the specialist in PC+ during P1 and P2. In both periods, the decision to refer to PC+ was based on GP consultation with the patient. The referral was first sent to the Transmural Interactive Patient Platform (TIPP), which accordingly planned and registered referrals to medical specialists (either in PC+ or outpatient hospital care). When patients were referred to PC+, they needed to have a recent X-ray (not older than six months) of the affected body part; if patients did not have a recent X-ray, they first went to the hospital to get one. In PC+, patients were seen by an orthopaedic surgeon or a senior resident in orthopaedic surgery of Maastricht UMC+ for a maximum of two consultations. In the PC+ centre, care is claimed as primary care performance so consultations are not subjected to the patient's deductible. Specialists treated patients and/or provided advice for GPs on further treatment strategies, and the GP retained responsibility for the patient.

If additional diagnostic tests were needed during P1, the referral decision following PC+ was automatic referral of the patient to outpatient hospital care. During P2, the specialist could request additional diagnostic tests within the PC+ setting. Patients had to visit the hospital only for these additional tests, and the test results determined whether a telephone consultation or follow-up consultation in PC+ was sufficient or whether a referral to outpatient hospital care for further diagnosis and/or treatment was necessary. An overview of the similarities and differences of P1 and P2 are shown in Table 1.



Note: P1 = period from January 2015 to December 2016; P2 = period from January 2017 to December 2017; TIPP = Transmural Interactive Patient Platform; PC+ = Primary Care Plus; GP = general practitioner

-- (dashed line) refers to optional steps in the PC+ process

Figure 1 Flowchart of the Primary Care Plus referral process.

Table 1 Requirements, possibilities and consequences of diagnostics in Primary Care Plus during period 1 and 2

	Period 1	**Period 2**
	January 2015–December 2016	January 2017–December 2017
Requirements for a consultation in PC+	Patients need to have a recent X-ray (not older than six months) of the affected body part	Patients need to have a recent X-ray (not older than six months) of the affected body part.
Additional diagnostic possibilities	No (or limited) possibility to request additional diagnostic tests (MRI & ultrasound)	Possibility to request additional diagnostic tests (MRI & ultrasound)
Consequences if additional diagnostics are needed	If additional diagnostics are required, patients are referred to outpatient hospital care	If additional diagnostics are required, patients are referred to outpatient hospital care for diagnostic purposes only. Follow-up consultations can take place at PC+

Note: PC+ = Primary Care Plus; MRI = magnetic resonance imaging

Data collection

In this study, the data of all patients visiting PC+ for orthopaedic care were collected. The data of the two independent PC+ centres were merged into one data set. These data consist of patient age, gender, final established diagnosis, number of consultations in PC+, treating specialist, and the referral decision following PC+. The patient diagnosis was labelled according to the diagnosis group classification list, which is part of the Diagnosis Treatment Combination (DBC) [30].

Outcome measures

The primary outcome of the present study was the referral decision following PC+ (back to the GP or referral to outpatient hospital care). Independent variables (or predictors) were consultation-related factors (number of consultations in PC+ and treating specialist) and patient-related factors (patient age, gender, and final established diagnosis). The treating specialist variable was divided into six categories; the first five categories included the five most common orthopaedic surgeons working in PC+, and the sixth category included all other orthopaedic surgeons working in PC+. Patient diagnoses were registered by the orthopaedic surgeon after the last consultation in PC+ according to the International Classification of Diseases (ICD-10) [31]. This variable was divided into 12 categories; the first 10

categories included the 10 most common diagnoses in PC+, an 11th category included all other diagnoses and a 12th category included all patients with a missing diagnosis. In addition, the number of MRIs and ultrasounds requested during P2 were described.

Statistical analysis

To examine the effect of the possibility to request diagnostic tests in PC+, referral decisions and consultation- and patient-related factors were compared between P1 and P2. Continuous data were presented as the mean and 95% confidence interval (CI) and were compared using an independent-sample *t*-test. Categorical data were presented as counts and percentages, and were compared using Pearson's x^2 test, with presenting the 95% CI for 2x2 tables. *P*-values ≤ 0.05 were considered statistically significant.

To describe variations in the referral decision, logistic regression modelling was used, with the decision to refer to outpatient hospital care being a binary yes/no variable. Firstly, univariate logistic regression analysis was used to evaluate the relationship between the referral decision following PC+ and the consultation- and patient-related predictors. Predictors with a *p*-value ≤ 0.25 were simultaneously entered into the multivariable model (enter method; [32]). Among the categorical predictors, treating specialist and patient diagnosis—the categories with the smallest difference in referral decision between P1 and P2—were selected as the reference group.

The effect of time (P2 vs. P1) on the difference in referral decisions between specialists as well as on the difference in referral decisions between diagnoses were analysed using specialist-period and diagnosis-period interactions, respectively. A significant interaction indicates that the effect of the treating specialist or the patient diagnosis on the referral decisions depends on the period. In this case, interpreting the effect of the individual predictors in isolation can be misleading [33]. The results of the logistic regression were presented as odds ratios (OR) with a 95% CI. P-values ≤0.05 were considered statistically significant. The explained variation in the regression model was measured by the Nagelkerke pseudo-R^2 [34]. All analyses were performed using the SPSS software for Windows, version 25.0 (SPSS Inc., Chicago, IL, USA).

Results of the analyses were discussed during an expert meeting with three involved orthopaedic surgeons. The purpose of this meeting was to verify the findings and to contribute to a better interpretation of the results.

RESULTS

During the total study period, from January 2015 to December 2017, 2,534 patients visited PC+ for orthopaedic care. The referral decision following PC+ for 96 patients was unknown, so these patients were excluded from the analysis. The remaining 2,438 patients had, in total, 2,766 consultations in PC+, with a mean of 1.13 (95% CI = 1.12-1.14) (Table 2). Following PC+, 67.2% (N = 1,638) of patients were referred back to their GP and 32.8% (N = 800) to outpatient hospital care for further treatment/examination.

During P1, 1,384 patients visited PC+ for orthopaedic care. In total, these patients had 1,507 consultations in PC+, with a mean of 1.09 (95% CI = 1.07-1.11). Following PC+, 60.3% (N = 834) of patients were referred back to their GP and 39.7% (N = 550) to outpatient hospital care for further treatment/examination.

During P2, 1,054 patients visited PC+ for orthopaedic care. In total, these patients had 1,259 consultations in PC+, with a mean of 1.19 (95% CI = 1.17-1.21). Following PC+, 76.3% (N = 804) of patients were referred back to their GP and 23.7% (N = 250) to outpatient hospital care for further treatment/examination.

When comparing both periods, significantly less patients were referred to outpatient hospital care during P2 (95% CI = 0.40-0.56) and patients had significantly more consultations in PC+ during this period (95% CI = -0.13 - -0.08). Finally, with respect to the treating specialist and patient diagnosis, there was a significant difference in the distribution between P1 and P2 ($p \leq 0.001$).

Diagnostic tests

During P2, specialists working in PC+ requested 174 MRIs and 57 ultrasounds. In total, 21.8% (N = 230) of all PC+ patients were referred for an additional diagnostic test.

Predictors of referral to outpatient hospital care

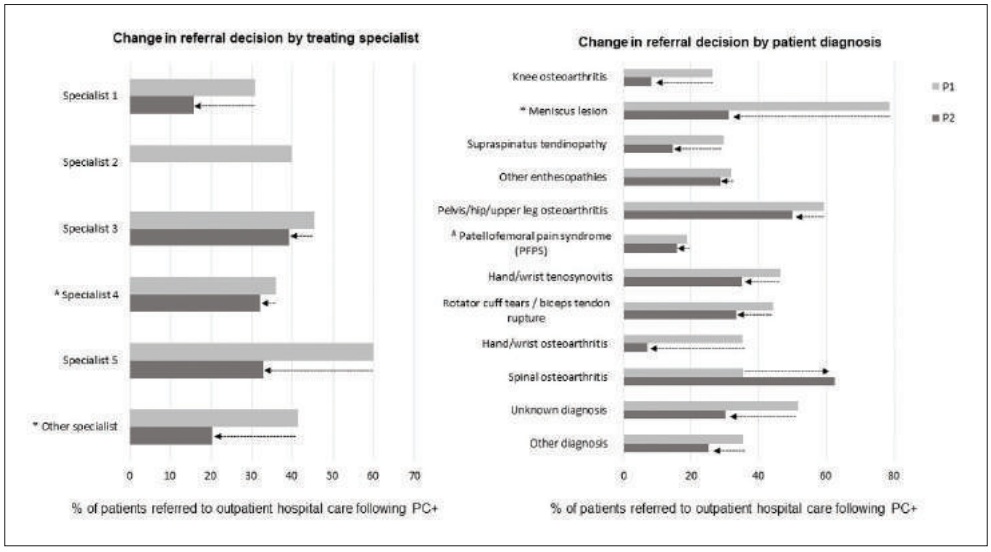
Specialist 4 and a diagnosis of patellofemoral pain syndrome (PFPS) were selected as the reference group for the logistic regression analysis, because they showed the least change in referral decision following PC+ when P1 and P2 were compared (Figure 2).

Univariate logistic regression analysis was performed to select potential predictors for the multivariable model. Based on the pre-set *p*-value criteria cut-off point of ≤ 0.25, all predictors, with the exception of gender and number of consultations, were included in the multivariable model (Table 3).

With respect to the multivariable model, it appeared that with increasing age, the likelihood of patients being referred to outpatient hospital care decreased significantly with OR = 0.86 (95% CI = 0.81-0.91) for every 10 years. Moreover, the multivariable model was adjusted for the possible confounding effect of the difference in age between patients in P1 and P2 (as described in Table 2).

The multivariable model, with the interaction terms included, showed a significant effect for the interaction between the treating specialist and period (p = 0.015) and between patient diagnosis and period (p ≤ 0.001; Table 3). Regarding the interactions between the treating specialist and the period, the period appeared to have a significantly different effect on the referral behaviour to secondary outpatient care for specialists from the category 'other specialist' (OR 0.40; 95% CI = 0.19 - 0.84) compared with the effect of the period on the referral behaviour of the reference group. As can be seen in Figure 2, Specialists from the category 'other specialist' showed a decrease in the number of referrals to oupatient hosptial care following PC+. Specialists 1 and 5 also showed a strong decrease in the number of referrals to outpatient hospital care (Figure 2), but this decrease was not significant compared with the reference group, which can be explained by the limited number of patients within those categories. The interactions between patient diagnosis and period showed a significantly different effect of the period on patients diagnosed with a meniscus lesion (OR 0.12; 95% CI = 0.03-0.45) compared with the effect of the period on the reference group. As can be seen in Figure 2, this diagnosis showed a strong decrease in the number of patients referred to outpatient hospital care following PC+.

Because of the significant interaction terms, it is not relevant to interpret the isolated effects of period, treating specialist and patient diagnosis. The multivariable model with interaction terms explained 17.6% of the variation (Nagelkerke $R^2 = 0.176$).



Note: P1 = period from January 2015 to December 2016; P2 = period from January 2017 to December 2017

** p ≤ 0.05; A Reference category in the multivariable model*

Figure 2 Change in referral decision following Primary Care Plus (period 1 versus period 2)

Table 2 Overview and comparison of Primary Care Plus patients and consultation characteristics during period 1 and 2

	Total (N = 2,438)	**P1 (N = 1,384)**	**P2 (N = 1,054)**
Referral decision % (N)			
Referral back to GP	67.2 (1,638)	60.3 (834	76.3 (804)
Referral to hospital care	32.8 (800)	39.7 (550)	23.7 (250)
Age in years mean (95% CI)	53.4 (52.7 - 54.1)	52.7 (51.8 - 53.6)	54.4 (53.4 - 55.4)
Gender % (N)			
Male	43.3 (1,056)	42.2 (584)	44.8 (472)
Female	56.7 (1,382)	57.8 (800)	55.2 (582)
Number of consultations mean (95% CI)	1.13 (1.12 - 1.14)	1.09 (1.07 - 1.11)	1.19 (1.17 - 1.21)
Treating specialist % (N)			
Specialist 1	13.0 (318)	14.7 (204)	10.8 (114)
Specialist 2	11.5 (281)	20.3 (281)	0.0 (0) [A]
Specialist 3	10.4 (253)	10.2 (141)	10.6 (112)
Specialist 4	8.1 (198)	10.0 (139)	5.6 (59)
Specialist 5	5.3(128)	1.8 (25)	9.8 (103)
Other specialist	51.7 (1,260)	43.0 (594)	63.6 (666)
Patient diagnosis % (N)			
Knee osteoarthritis	14.1 (344)	11.8 (164)	17.1 (180)
Meniscus lesion	7.0 (171)	8.2 (113)	5.5 (58)
Supraspinatus tendinopathy	6.8 (167)	6.6 (91)	7.2 (76)
Other enthesopathies	6.2 (150)	6.8 (94)	5.3 (56)
Pelvis/hip/upper leg osteoarthritis	4.5 (110)	4.6 (64)	4.4 (46)
Patellofemoral pain syndrome (PFPS)	4.0 (97)	3.8 (53)	4.2 (44)
Hand/wrist tenosynovitis	3.3 (80)	3.1 (43)	3.5 (37)
Rotator cuff tears/biceps tendon rupture	2.6 (63)	2.6 (36)	2.6 (27)
Hand/wrist osteoarthritis	2.6 (63)	2.5 (34)	2.8 (29)
Spinal osteoarthritis	2.3 (56)	3.5 (48)	0.8 (8)
Unknown diagnosis	10.8 (264)	9.0 (124)	13.3 (140)
Other diagnosis	35.8 (873)	37.6 (520)	33.5 (353)

Note: GP = general practitioner; CI = confidence interval; PC+ = Primary Care Plus; P1 = period 1; P2 = period 2

** $p \leq 0.05$; [A] Specialist did not work at PC+ during this period*

	Difference between P1 and P2	
	95% CI	*p*-values
	0.40 – 0.56	≤ 0.001*
	-3.07 – -0.35	0.014*
	0.95 – 1.31	0.202
	-0.13 – -0.08	≤ 0.001*
	-	≤ 0.001*
	-	≤ 0.001*

Table 3 Logistic regression analysis for referral to outpatient hospital care among orthopaedic patients in Primary Care Plus (N = 2,438)

Predictor	Univariate model	
	OR (95% CI)	***p*-value**
Age [A]	0.86 (0.82, 0.91)	≤.001*
Gender(male)	1.00 (0.85, 1.19)	0.966
Number of consultations	0.89 (0.69, 1.14)	0.343
Period (P2)	0.47 (0.40, 0.56)	≤.001*
Treating Specialist		
Specialist 1	0.64 (0.43, 0.94)	0.023*
Specialist 2	1.39 (0.95, 2.05)	0.091*
Specialist 3	1.24 (0.85, 1.81)	0.266
Specialist 4	...[B]	
Specialist 5	1.16 (0.73, 1.84)	0.529
Other specialist	0.81 (0.59, 1.11)	0.192*
Patient diagnosis		
Knee osteoarthritis	0.95 (0.53, 1.73)	0.878
Meniscus lesion	7.87 (4.28, 14.45)	≤.001*
Supraspinatus tendinopathy	1.39 (0.73, 2.62)	0.314
Other enthesopathies	2.08 (1.11, 3.90)	0.022*
Pelvis/hip/upper leg osteoarthritis	5.86 (3.08, 11.16)	≤.001*
Patellofemoral pain syndrome (PFPS)	...[B]	
Hand/wrist tenosynovitis	3.30 (1.66, 6.57)	0.001*
Rotator cuff tears/biceps tendon rupture	3.10 (1.50, 6.41)	0.002*
Hand/wrist osteoarthritis	1.35 (0.61, 2.97)	0.464
Spinal osteoarthritis	3.05 (1.44, 6.44)	0.004*
Unknown diagnosis	3.16 (1.77, 5.63)	≤.001*
Other diagnosis	2.14 (1.24, 3.68)	0.006*

Note: OR= odds ratio; CI = confidence interval

** p ‹ 0.25 (univariate analysis); ** p ‹ 0.05 (multivariable analysis); [A] Age was rescaled such that one unit is equal to 10 years; [B] Reference category in the multivariable model; [C] Variable not included in the multivariable model*

Multivariable model with interaction terms	
OR (95% CI)	***p*-value**
0.86 (0.81, 0.91)	≤0.001**
…[C]	
…[C]	
1.72 (0.48, 6.21)	0.409
0.87 (0.53, 1.41)	0.561
1.32 (0.77, 2.24)	0.312
1.34 (0.85, 2.11)	0.206
…[B]	
2.76 (1.08, 7.05)	0.034**
1.42 (0.94, 2.15)	0.098
2.34 (1.06, 5.20)	0.037**
19.43 (8.42, 44.83)	≤0.001**
2.51 (1.08, 5.82)	0.032**
2.62 (1.14, 6.01)	0.023**
10.05 (4.17, 24.25)	≤0.001**
…[B]	
5.63 (2.21, 14.33)	≤0.001**
5.67 (2.13, 15.10)	0.001**
3.25 (1.19, 8.90)	0.022**
3.30 (1.30, 8.37)	0.012**
6.02 (2.68, 13.52)	≤0.001**
2.91 (1.41, 6.00)	0.004**

Table 3 Continued

Predictor	Univariate model	
	OR (95% CI)	***p*-value**
Interaction specialist x period		
Specialist 1	-	-
Specialist 2	-	-
Specialist 3	-	-
Specialist 4	-	-
Specialist 5	-	-
Other specialist	-	-
Interaction diagnosis x period		
Knee osteoarthritis	-	-
Meniscus lesion	-	-
Supraspinatus tendinopathy	-	-
Other enthesopathies	-	-
Pelvis/hip/upper leg osteoarthritis	-	-
Patellofemoral pain syndrome (PFPS)	-	-
Hand/wrist tenosynovitis	-	-
Rotator cuff tears/biceps tendon rupture	-	-
Hand/wrist osteoarthritis	-	-
Spinal osteoarthritis	-	-
Unknown diagnosis	-	-
Other diagnosis	-	-

Note: OR= odds ratio; CI = confidence interval

** p ‹ 0.25 (univariate analysis); ** p ‹ 0.05 (multivariable analysis); A Age was rescaled such that one unit is equal to 10 years; B Reference category in the multivariable model; C Variable not included in the multivariable model*

	Multivariable model with interaction terms	
	OR (95% CI)	***p*-value**
		0.015**
	0.43 (0.17, 1.09)	0.076
	-	-
	1.05 (0.41, 2.71)	0.924
	...[B]	
	0.39 (0.12, 1.28)	0.120
	0.40 (0.19, 0.84)	0.015**
		≤0.001**
	0.29 (0.08, 1.01)	0.052
	0.12 (0.03, 0.45)	0.002**
	0.46 (0.12, 1.75)	0.253
	1.10 (0.30, 4.04)	0.890
	0.66 (0.18, 2.50)	0.541
	...[B]	
	0.60 (0.15, 2.47)	0.479
	0.70 (0.15, 3.14)	0.637
	0.18 (0.03, 1.24)	0.081
	4.86 (0.72, 32.73)	0.104
	0.33 (0.10, 1.17)	0.086
	0.74 (0.24, 2.26)	0.597

DISCUSSION

The present study evaluated referral decisions following orthopaedic care in PC+, taking into account the influence of the availability of diagnostic tests on the referral decision, as well as consultation- and patient-related predictors of these decisions.

The apparent influence of the possibility to request additional diagnostic tests on the referral decision is in accordance with the study by van Hoof et al. [28]. In the present study, specialists indicated that they needed diagnostic imaging, such as an X-ray, to diagnose patients. During P1 in the current setting of PC+, orthopaedic surgeons required a recent X-ray for all patients prior to the first consultation. Orthopaedic surgeons mentioned that, based on previous experience, approximately 70% of patients would have to obtain an X-ray following the first consultation, so not mandating an X-ray beforehand would make PC+ less effective. Referral rates to outpatient hospital care during this period were, according to the involved stakeholders, still considerable due to the need for additional diagnostic tests. To increase the effectiveness of orthopaedic care in PC+, stakeholders decided to introduce the possibility of requesting additional diagnostic tests, such as MRIs and ultrasounds. As a result, the number of referrals to outpatient hospital care decreased significantly during P2. Despite these positive results, it is important to be wary of unnecessary care in PC+. Because the initial aim of PC+ was to limit the availability of diagnostic tools to promote the generalist approach, it is necessary to determine to what extent diagnostic tests fit within this vision. This is also relevant for ensuring cost-effectiveness and patient-centered care. The number of consultations in PC+ also increased significantly, which can be explained by the fact that specialists needed an extra consultation to discuss the test results with the patient.

Older patients were less likely to be referred to outpatient hospital care following PC+. Similar findings were also reported by McBride et al. [35] in a study on referral variation from primary to secondary care of patients with, among other ailments, hip pain. Possible explanations given in this study were patient preferences and the clinical uncertainty regarding the benefits and adverse effects of treatment for elderly patients [35-37]. Although older patients are slightly more at risk following hip or knee surgery, for example, the quality of life can also increase in this group [38, 39]. In addition, Becker et al. [40] found that younger patients with hand osteoarthritis had a greater likelihood of surgery and also had increased healthcare-related costs.

All specialists showed a decrease in the number of referrals to outpatient hospital care during P2. From the perspective of substituting primary care for specialised medical care, this was the desired effect; however, the extent to which it is desirable to change the referral behaviour of specialists when more diagnostic tests are available is questionable. Based on the existing literature and the previous discussion of the extent to which diagnostic tests fit within the vision of PC+, it is important to remain critical towards the availability of diagnostic tests to ensure overuse is not encouraged [41, 42]. According to Vierhout et al. [24], diagnostic tests carried out by the orthopaedic suregeon are not always needed and they might be requested based on routine. The availability of diagnostic tests in PC+ should therefore be a topic for discussion among the involved stakeholders, and the awareness among specialists regarding the necessity of diagnostic tests should be enhanced [43].

The novelty of the PC+ setting also requires specialists to deal with a new context and environment as well as their related expectations and understandings of what best practice is in this setting [44]. PC+ involves more than shifting outpatient hospital care to the primary setting; it is also about changing the mind-sets and behaviour of the involved health care professionals—both the GPs and medical specialists [45]. As described by Gupta et al. [44], changing clinical practice is a complex process of learning and unlearning. The degree to which medical specialists succeed in changing their behaviour according to the PC+ setting is questionable. Based on our findings about PC+, practice patterns varied among the involved orthopaedic surgeons. More research is therefore needed to specify relevant features of medical specialists to work in PC+, taking into account the specific setting of PC+. The number of medical specialists working in PC+ is a related area for discussion. In total, 37 different specialists (orthopaedic surgeons and specialty trainees) worked in PC+ during the study period. This number indicates a high turnover of specialists, which could be a barrier to the development of practice patterns in this new setting and could limit any possible learning effect between specialists and GPs or the opportunity to overcome the knowledge gap in primary care [19]. Stimulating collaboration between medical specialists and GPs may be associated with improved health outcomes, optimised care, and less use of hospital care [46], so assigning a select group of appropriate specialists for PC+ could improve the effectivenes of the program.

Regarding patient diagnosis, several diagnoses showed a decrease in referrals to secondary care during P2, but patients diagnosed with spinal osteoarthiritis showed an increase in referrals to secondary care during P2, which indicates that spinal

osteoarthritis appears to be less appropriate for PC+ even when additional diagnostic tests are available. During the expert meeting, specialists confirmed this assumption. Patients with back problems should not be referred to PC+ for orthopaedic care, but should be referred to a specific back pain clinic. Development of patient profiles for PC+ appears relevant for further optimisation of the program. These profiles can give an indication of patient complaints that are suitable for PC+, which would support GPs in their PC+ referral behaviour [28].

LIMITATIONS

The variation of 17.6% explained by the final model in the present study suggests that many factors influence referral decisions following PC+. The number of predictors included in the present study was restricted, which was inherent to the use of monitoring data containing a limited amount of information. More information, such as the International Classification of Primary Care (ICPC) codes [47]. and registration of the severity of the complaint, would likely lead to a better prediction of the referral decision and therefore be more recognisable and manageable for GPs. Accordingly, appropriate referrals to PC+ will increase, and consequentially so will the intervention's efficiency.

Moreover, a large number of different diagnoses were determined by the specialists in PC+ (N = 138). Only the 10 most common diagnoses were divided in separate categories, accounting for 53% of all consultations. The remaining diagnoses (N = 128) were merged into an 11th category, accounting for 36% of all consultations. Additionally, the diagnoses of 264 patients (11%) were missing, which was partly caused by the specialists becoming accustomed to the registration method at the beginning of PC+, but this is not supposed to influence the results considerably.

CONCLUSIONS

The results of this study reveal that the possibility of requesting additional diagnostic tests for orthopaedic surgeons working in a primary care setting significantly decreased the number of referrals to secondary outpatient care. With more than three-quarter of the patients referred back to their GP during P2, orthopaedic care seems to fit to the aim of PC+ to prevent unnecessary referrals to hospital care. However, more research is needed regarding the effectiveness and suitability of the use of diagnostic tests to further optimise orthopaedic care in PC+. Selection of the

appropriate profiles to indicate suitable specialists and patients for PC+ is therefore recommended, because both significantly influenced the referral decision. Other factors such as volume, planning and duration of consultations, quality of care, patient health status, and cost of care should also be taken into account in future research to further optimise the substitution of orthopaedic care and reduce rising healthcare costs.

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04

Does the implementation of a care pathway for patients with hip or knee osteoarthritis lead to fewer diagnostic imaging and referrals by general practitioners?

A pre-post-implementation study of claims data

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ABSTRACT

Background: The Dutch care for hip and knee osteoarthritis (OA) is of good quality, but there is room for improvement regarding the efficient use of diagnostic imaging and conservative treatment. Therefore a stepped-care approach, in the shape of the care pathway 'Better exercise in osteoarthritis', was implemented to reduce the number of diagnostic imaging requested by GPs and referrals of GPs to orthopaedic care.

Methods: In 2015, the pathway is implemented with the use of educational meetings, distributing guidelines and incorporating reminders in the GPs' referral application. To evaluate the effect of the pathway on the diagnostic and referral behaviour of GPs, hip and knee related health insurance claims are used together with claims of other joints and of a control region for comparison. The average number of claims and the percentage change in the post-implementation period are described. Binary logistic regression analysis is used to examine the interaction between region (intervention and control) and period (pre- and post-implementation). Using random sampling of patient records, information about the practical application of the pathway and the number of hip or knee arthroplasties is added.

Results: In both regions, the number of diagnostic imaging decreased and the number of initial orthopaedic consultations increased during the post-implementation period. Significant interaction effects were found in knee-related diagnostics ($p \leq 0.001$) and diagnostics of other joints ($p = 0.039$). No significant interaction effects were found in hip-related diagnostics ($p = 0.060$) and in initial orthopaedic consultation claims of hip ($p = 0.979$), knee ($p = 0.281$), and other joints ($p = 0.464$). Being referred according to the pathway had no significant effect on the probability of undergoing arthroplasty.

Conclusion: The implementation of the pathway had a positive effect on GPs diagnostic behaviour related to the knee, but not to the hip. The referral behaviour of GPs to orthopaedic care needs attention for future interventions and research, since an increase (instead of a desired decrease) in the number of initial orthopaedic consultations was found. Focusing on the entire width of care for hip and knee OA could be helpful.

BACKGROUND

Osteoarthritis (OA) is a common joint disorder affecting more than half of the population aged 65 years and older [1, 2]. This long-term chronic disease is often associated with stiffness, pain, and functional limitations [3, 4].Together, this results in a significant reduction in the quality of life of these patients [5, 6].

In 2016, an estimated 1.25 million people (around 7% of the population) had the diagnoses OA in the Netherlands [7]. Annually, there are approximately 140,000 new cases of OA in the country. Knee OA is most common, followed by hip OA. Based on demographic trends, it is expected that the number of people with OA will increase by almost 41% between 2015 and 2040 [7]. Recent increases in the number of people with obesity, a major determinant of OA, suggests that the prevalence of OA is likely to rise in future [8-10]. In 2015, 1.3% of the total cost of health care in the Netherlands was spent on OA-related care [11]. In view of the increasing prevalence, these costs are likely to rise substantially.

In 2014, the Dutch National Health Care Institute (in Dutch: Zorginstituut Nederland) stated that the care for hip and knee OA in the Netherlands is of good quality, but it also emphasised that there is room for improvement in some areas [12]. One of these suggested improvements is related to the efficient use of diagnostic imaging, such as X-ray or magnetic resonance imaging (MRI). OA is primarily a clinical diagnosis [13]. This implies that in most cases the diagnosis can be based on history taking and physical examination [14, 15]. Despite the recommendations in the guidelines [15], Smink et al. [16] found that general practitioners (GPs) often request for diagnostic imaging.

Another suggestion for improvement is related to the treatment of OA. International evidence-based guidelines for hip and knee OA recommend starting with non-surgical (conservative) treatments [17-21]. Joint replacement surgery (arthroplasty) should be performed only in advanced OA and not in the early stages, given the limited lifespan of prostheses and the less successful outcomes of revision arthroplasty [22-24].

Despite the availability of guidelines, several studies have found that a majority of the patients referred to an orthopaedic surgeon did not receive appropriate prior conservative treatment [25-29]. This can be explained by the lack of practical and clear recommendations and strategies about the necessity and sequence of treatment options [30]. A systematic and period approach, a so-called ‘stepped care’ strategy, can be a tool to optimise the use of existing conservative treatment options

[31, 32]. Stepped care is characterised by interventions that are offered not earlier or with more intensity than necessary. More radical interventions, like hip or knee arthroplasty, should only be considered when patients do not respond sufficiently to conservative treatment options [27, 33].

An example of a stepped care approach is the care pathway 'Better exercise in osteoarthritis' (in Dutch: 'Beter bewegen bij artrose') implemented in the Western Mining District of Limburg, in the South of the Netherlands [34]. Various stakeholders, like GPs, physiotherapists, and an orthopaedic surgeon, are involved in this intervention. The pathway is based on the guideline of the Dutch College of General Practitioners (NHG) [35] and aims to treat patients with knee or hip OA according to a stepped care approach. Furthermore, this pathway clearly states that OA is a clinical diagnosis, and therefore X-rays are not necessary.

The pathway may positively influence quality of care and health outcomes. In addition, unnecessary costs could be avoided by implementing these improvements [12]. The Dutch National Health Care Institute estimated that 90%of the costs associated with diagnostic imaging related to both hip and knee OA are unnecessary and that with the implementation of the guidelines, more than €14 million could be saved by deploying conservative treatment [12]. In addition, 5% of hip arthroplasties and 10% of knee arthroplasties could be prevented, based on the assumption that a group of patients is already appropriately managed in primary care. This could result in an additional saving of €34 million [12].

This study aims to evaluate the effect of the implementation of the pathway, on GP diagnostic imaging requests and GP referrals to orthopaedic surgeons for hip and knee OA. In addition, this study evaluates to what extent the pathway is applied in practice before patients are referred to orthopaedic care and the effect of the pathway on the appropriateness of these referrals.

METHODS

Design

This is an observational study comparing the diagnostic and referral behaviour of GPs in the pre- and post-implementation period of the intervention using health insurance claims data from 2012 to 2016. In addition, a patient record review is conducted

to get more information about the practical application of the pathway and the appropriateness of referrals to orthopaedic care within the intervention region.

Setting

The pathway originates from a regional collaboration of stakeholders in the Western Mining District located in the South of Limburg. Stakeholders consist of a coordination centre for diagnostics, MCC Omnes; the GP organisation Meditta; Zuyderland Medical Centre (MC) (location Sittard-Geleen); the health insurance company CZ; and a patient representative organisation, Citizen Power (in Dutch: Burgerkracht). These organisations work together to provide the right care in the right place [34].

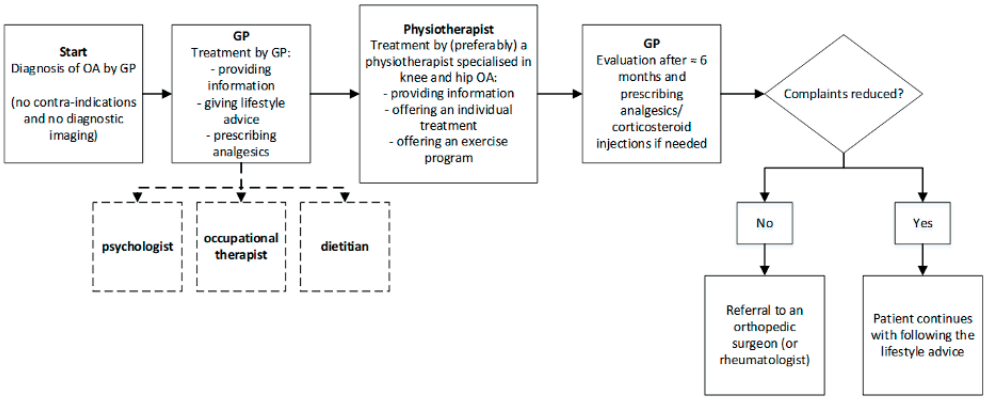
The Western Mining District has a population of about 185,000 people. The population is declining and ageing [36]. The control group incorporated three other regions. These regions are selected because they are located in the same province as the intervention region and are also characterised by a declining and ageing population. Together, these control regions have a population of about 690,000 people.

In the Netherlands, having health insurance is mandatory [37]. In the Western Mining District, CZ is the health insurance company with the largest market share in the region. In addition, all Dutch residents are registered at a GP practice. Primary care is delivered by GPs, which initiates diagnostics and acts as a gatekeeper to specialised medical care [38]. GP consultations are fully covered by the health insurance [39]. For consulting a medical specialist, a yearly compulsory deductible is levied. This implies that there is a certain amount of specialised medical treatment expenses that a patient has to pay out of pocket before the health insurance company will compensate the expenses. The same applies for diagnostic tests (including diagnostic imaging) and pharmaceuticals prescribed by GPs. The amount of the deductible is determined by the Dutch government and changes every year [40]. During the study period (2012-2016), the amount increased from €220 to €385.

Intervention

The pathway is designed, using the national guidelines for hip and knee OA [34], by members of an expert group, consisting of two GPs, a physical therapist, an orthopaedic surgeon, a rheumatologist, a radiologist, a physician assistant and a coordinator of MCC Omnes. In February 2015, the pathway is implemented in the Western Mining District based on three interventions: educational meetings, distribution of the guidelines, and reminders. All interventions are developed and

coordinated by members of the expert group and focus on improving the stepped care approach of hip and knee OA treatment, reducing diagnostic imaging requests and reducing referrals to orthopaedic surgeons. The educational meetings consist of one meeting organised for GPs and physiotherapists together at the start of the implementation process, followed by advanced educational courses organised separately for GPs and physiotherapists. The educational meetings focus on



Note: OA = osteoarthritis; GP = general practitioner
- - (dashed line) refers to optional steps in the care pathway

Figure 1 Process of the stepped care approach in the pathway

recognising OA and red flag situations, the content of the pathway and related guidelines, the role of different professionals within the pathway (with emphasis on GPs and physiotherapists) and the application of the pathway in practice, for example by discussing practical cases, patient communication skills training and practicing administering corticosteroid injections. The educational meetings are voluntary and professionals earn medical education credits for their presence. Around 20% of the GPs in the intervention region attended the first education meeting. In addition, the expert group assumes that the attended GPs spread the content of the meetings among their colleagues within their general practice and that all GPs, affiliated with MCC Omnes, eventually conform to their initiatives. To further support the dissemination of the pathway, a visualisation and explanation of the stepped-care approach are placed at the website of MCC Omnes, and in the newsletter and on the mobile application of MCC Omnes to reach all GPs in the region. In addition, to support GPs in applying the pathway in practice, a reminder pops up in the GPs' referral application (called ZorgDomein) when requesting hip or

knee related diagnostic imaging or when referring patients with hip or knee related complaints to orthopaedic care. This reminder forces GPs to indicate which steps of the pathway have been followed prior to the request or referral. Hence, all GPs are informed about the pathway through these different channels. The pathway focusses on patients with hip and knee OA diagnosed by their GP based on history taking and physical examination. In cases of rheumatic diseases, previous diagnosis of OA, OA that cannot be explained sufficiently, young age (‹45 years), prominent polyarthritis (in multiple joints), (familial) psoriasis, or inflammatory bowel disease, patients are excluded from being treated according to the pathway.

Figure 1 shows the stepped care process of the pathway. When patients are diagnosed with OA by their GP and patients are eligible for treatment according to the pathway, the GP provides information about OA and advice about lifestyle. When necessary, analgesics are prescribed. The GP refers the patient, according to the conservative policy, to a physiotherapist and when necessary to a dietitian, psychologist (in case of problems with coping), and/or occupational therapist. The physiotherapist provides more (tailored) information about OA to the patient. In addition, an individual treatment process is started, aimed at reducing pain and functional disorders, combined with an exercise program focussing on guiding patients to a more active lifestyle. Preferably, after approximately six months, the GP evaluates the results of the conservative treatment. When complaints reduce, patients are advised to continue with the lifestyle advice. When complaints not reduce, additional analgesics could be prescribed or corticosteroid injections are administered. Finally, patients could be referred to specialised medical care (mostly a referral to an orthopaedic surgeon).

Data collection

OBSERVATIONAL STUDY

The effect of the pathway is assessed using annual health insurance claims data (2012 to 2016) from the health insurance company CZ. Claims data can be categorised as health care administrative data [41]. The health insurance company collects these data for administrative and billing purposes, and they can be used to study health care consumption. The claims data are based on measurements for all CZ insured persons within the intervention and control region (e.g. census data).

The number of hip- and knee-related claims are compared with claims related to other joints (neck, shoulder, upper arm, elbow, forearm, hand, wrist, fingers, ankle, foot,

and toes). Additionally, the number of health insurance claims in the Western Mining District (intervention region) are compared with the number of claims in the control region.

PATIENT RECORD REVIEW

To collect information about the practical application of the pathway and the appropriateness of referrals to orthopaedic care within the intervention region, a patient record review is conducted using random sampling. Approximately 10% of the total number of the records of patients with a suspicion of hip or knee OA, who were referred for an initial orthopaedic consultation during the period from February 2015 to October 2016 (post-implementation period), are included.

Outcome measures

OBSERVATIONAL STUDY

Regarding the claims data, the primary outcome measures are the number of hip- and knee-related diagnostic imaging procedures (X-rays and MRIs) requested by GPs and the number of GP referrals to orthopaedic care per 1,000 insured persons.

Based on the claims data, it is not possible to determine the total number of GP referrals to orthopaedic care, due to a lack of follow-up of the referral or non-attendance [42]. Therefore, the actual claims of initial orthopaedic consultations related to OA of the hip and knee and other joints are used, both in the intervention and control region. Initial consultations are recognised by the so-called Diagnosis Treatment Combination (DTC) (in Dutch: Diagnose Behandel Combinatie, DBC) [43]. Every DTC has a unique performance code that includes all information about the type of care (initial or follow-up), the demand for care, the diagnosis and type of treatment.

The characteristics of the entire population insured by CZ in the intervention region and the control region are 1) the number of insured persons, presented with averages per period, 2) gender distribution, presented using the percentages of men, and 3) average age, presented with means and standard deviations (SDs).

The pre- and post-implementation period is determined by the implementation of the pathway in the beginning of 2015. Therefore, the pre-implementation period is determined as January 2012 to December 2014 and the post-implementation period is determined as January 2015 to December 2016. A post-implementation period of two years is selected. It is not possible to select a period of three years due to the delay in the processing of the claims data by the health insurer.

Patient record review

The collected patient records using the random sampling contain the answers to the questions related to the reminder that pops-up in the GPs' referral application when they refer patients to orthopaedic care ("Did you (the GP) went through the steps of the pathway?"). The answers to this question (yes or no).The answers to this question (yes or no) are used to check whether patients had been referred according to the pathway. Furthermore, the patients' records contain information about the follow-up orthopaedic care in the hospital. Information about the diagnosis (yes/no OA) and the treatment (yes/no arthroplasty) are collected from these records to check if the referral was appropriate.

Analysis

OBSERVATIONAL STUDY

Characteristics of the intervention and control region during the pre- and post-implementation period are presented. In addition, the average number of health insurance claims for diagnostic imaging and initial orthopaedic consultations (separately for hip, knee, and other joints) per 1,000 insured persons in the pre- and post-implementation period of both regions are reported. The percentage change in the number of requested diagnostic imaging and initial orthopaedic consultations in the post-implementation period compared to the pre-implementation period is calculated and presented. Furthermore, the proportion of diagnostic imaging and initial orthopaedic consultations claims per 1,000 insured persons per region and per period are dichotomised to a binary variable (yes/no-claimed diagnostic imaging or initial orthopaedic consultation).

Binary logistic regression analyses are used to determine the influence of the implementation of the pathway on the proportion of health insurance claims for diagnostic imaging and initial orthopaedic consultations per 1,000 insured persons in the intervention region compared to the control region. The dependent variable in these models is the binary variable indicating claimed diagnostic imaging (yes/no) or claimed initial orthopaedic consultation (yes/no). The independent variables are region (intervention or control region) and period (pre- or post-implementation period) and the interactions between those variables, using the Enter method [44]. In addition, odds ratios (OR), p-values, and 95% confidence intervals (CI) are reported.

The number and percentage of patients referred to orthopaedic care according to the pathway, the number and percentage of patients diagnosed with OA as determined by the orthopaedic surgeon and the number and percentage of arthroplasties performed are presented in flow charts, separately for patients with a suspicion of hip and knee OA. Pearson's chi-square test is used to test the difference in the probability of undergoing arthroplasty between patients who were referred according to the pathway and patients who were not, again separately for patients with hip and knee OA.

Descriptive statistics and analyses are performed using SPSS version 25, and statistical significance is defined as $p < 0.05$ (IBM SPSS Statistics, Armonk, NY).

Expert meetings

The process of the pathway and results of the analyses are discussed during meetings with the expert group. The purpose of these meeting is to verify the findings and to contribute to a better interpretation of the results.

RESULTS

Observational study

Table 1 shows that the number of insured persons decreased over time in the intervention region. In the control region, the number of insured persons increased. However, the proportions of men and the mean age remained stable over time in both regions. Therefore, it is assumed that the effect of the decrease in the intervention region is limited.

The number of claims and the percentage change in the intervention and control region are described (Table 2). In both regions, the average number of requested diagnostic imaging procedures decreased during the post-implementation period. Regarding the initial orthopaedic consultations, an increase of claims during the post-implementation period in both regions is visible.

As presented in Table 3, there was no statistically significant difference in the decrease of the number of GP-requested hip-related diagnostic imaging during the post-implementation period (OR = 0.903, 95% CI = 0.812–1.004, p = 0.060) in the intervention region compared to the control region. However, during the post-implementation period the number of GP-requested knee-related diagnostic imaging

(OR = 0.781, 95% CI = 0.693–0.880, p ≤ 0.001) and GP-requested diagnostic imaging of other joints (OR = 0.931, 95% CI = 0.870–0.997, p = 0.039) declined statistically significantly more in the intervention region.

Moreover, there was no statistically significant difference in the increase of the number of initial orthopaedic consultation claims of the hip (OR = 1.002, 95% CI = 0.871–1.152, p = 0.979), knee (OR = 0.894, 95% CI = 0.728–1.097, p = 0.281), or other joints (OR = 1.091, 95% CI = 0.864–1.378, p = 0.464) in the intervention region compared to the control region (Table 4).

Table 1 Characteristics of intervention and control region

Region and period	Average number of insured persons (N)	Gender - male (%)
Intervention region		
Pre	85749	48.3
Post	80078	48.4
Control region		
Pre	295796	48.9
Post	299306	48.8

Note: SD = standard deviation

Table 2 Number of claims per region in the pre- and post-implementation period

	Requested diagnostic imaging		
	Pre	Post	
	Average per 1000 insured persons (N)	Average per 1000 insured persons (N)	Percentage change (%)*
Intervention region			
Hip-related	15.19	13.45	– 11.5
Knee-related	12.72	9.51	– 25.2
Other joints	35.45	32.14	– 9.3
Control region			
Hip-related	15.94	15.43	– 3.2
Knee-related	13.24	12.91	– 2.5
Other joints	43.95	43.73	– 0.5

** – = percentage change is negative (a decrease), and + = percentage change is positive (an increase)*

	Age in years (mean ± SD)		
	45.92 ± 23.26		
	46.36 ± 23.40		
	45.32 ± 23.26		
	45.78 ± 23.39		

	Initial orthopaedic consultation		
	Pre	Post	
	Average per 1000 insured persons (N)	Average per 1000 insured persons (N)	Percentage change (%)*
	10.92	13.14	+ 20.3
	5.93	6.47	+ 9.1
	3.07	3.60	+ 17.3
	11.30	13.50	+ 19.5
	5.37	6.77	+ 26.1
	5.04	5.34	+ 6.0

Table 3 Results of the logistic regression analysis for claimed diagnostic imaging

	Odds Ratio	P-value
Hip-related		
Period	0.981	0.573...
Region	0.895	0.003 *
Region x period	0.903	0.060...
Knee-related		
Period	0.955	0.225...
Region	0.898	0.008 *
Region x period	0.781	0.000 *
Other joints		
Period	0.970	0.152...
Region	0.764	0.000 *
Region x period	0.931	0.039 *

** $p \leq 0.05$*

Table 4 Results of the logistic regression analysis for initial orthopaedic consultation claims

	Odds Ratio	P-value
Hip-related		
Period	1.103	0.039 *
Region	0.939	0.222...
Region x period	1.002	0.979...
Knee-related		
Period	1.112	0.128...
Region	1.015	0.844...
Region x period	0.894	0.281...
Other joints		
Period	1.007	0.922...
Region	0.600	0.000 *
Region x period	1.091	0.464...

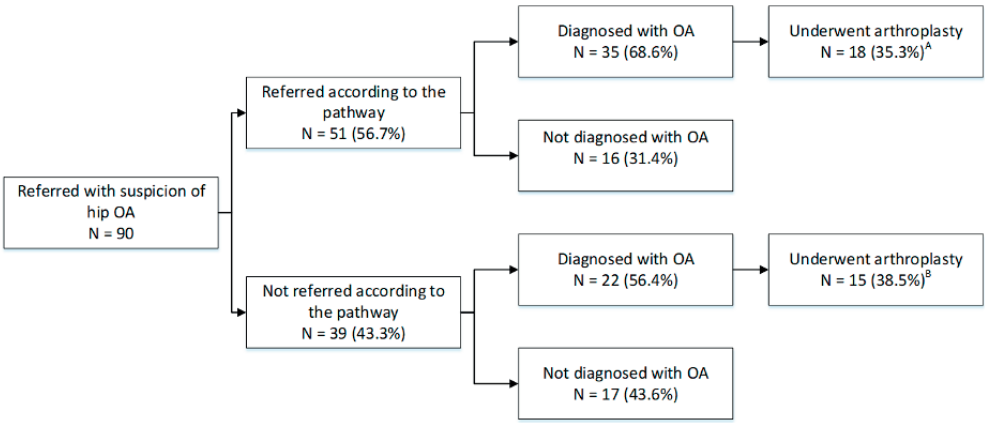
** $p \leq 0.05$*

	95% Confidence Interval	
	Lower Bound	Upper Bound
	0.916	1.050
	0.832	0.963
	0.812	1.004
	0.886	1.029
	0.829	0.973
	0.693	0.880
	0.930	1.011
	0.729	0.801
	0.870	0.997

	95% Confidence Interval	
	Lower Bound	Upper Bound
	1.005	1.209
	0.849	1.039
	0.871	1.152
	0.970	1.274
	0.877	1.173
	0.728	1.097
	0.878	1.155
	0.509	0.709
	0.864	1.378

Patient record review

Figure 2 and 3 show the number and percentage of patients referred by their GP (from the intervention region) to orthopaedic care with a suspicion of hip or knee OA and the number of performed arthroplasties. Figure 2 shows that the majority of patients with a suspicion of hip OA (56.7%) were referred according to the pathway. After the referral, 68.6% of the patients who were referred according to the pathway were diagnosed with OA. For patients not referred according to the pathway, this percentage was 56.4%. Finally, the percentage of patients who underwent arthroplasty was lower for patients referred according to the pathway than for patients not referred according to the pathway (35.3% and 38.5%, respectively). In addition, Figure 3 shows that the majority of patients with a suspicion of knee OA (53.8%) were referred according to the pathway. After the referral, 75.8% of the patients who were referred according to the pathway were diagnosed with OA. For patients not referred according to the pathway, this percentage was 59.0%. Finally, the percentage of patients who underwent arthroplasty was higher for patients referred according to the pathway than for patients not referred according to the pathway (39.6% and 28.2%, respectively).

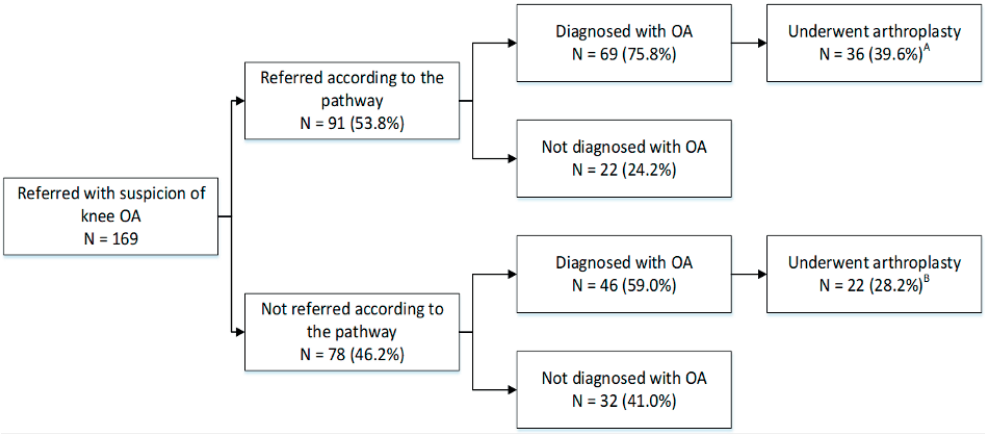


Note: OA = osteoarthritis

[A] Percentages are calculated based on the number of patients referred according to the pathway (N = 51)

[B] Percentages are calculated based on the number of patients not referred according to the pathway (N = 39)

Figure 2 Flow chart of the probability of arthroplasty for patients referred with suspicion of hip osteoarthritis



Note: OA = osteoarthritis
[A] Percentages are calculated based on the number of patients referred according to the pathway (N = 91
[B] Percentages are calculated based on the number of patients not referred according to the pathway (N = 78)

Figure 3 Flow chart of the probability of arthroplasty for patients referred with suspicion of knee osteoarthritis

Table 5 shows that for both patients with a suspicion of hip and knee OA, the probability of undergoing arthroplasty does not significantly differ for patients who were or were not referred according to the pathway.

Table 5 Percentages and numbers of arthroplasty in patients being referred or not according to the pathway

	Total N	Arthroplasty % (N)		P-value [A]
		Yes	No	
Hip OA	90			0.757
Referred according to the pathway	51	35.3 (18)	64.7 (33)	
Not referred according to the pathway	39	38.5 (15)	61.5 (24)	
Knee OA	169			0.121
Referred according to the pathway	91	39.6 (36)	60.4 (55)	
Not referred according to the pathway	78	28.2 (22)	71.8 (56)	

Note: OA = osteoarthritis

[A] Tested with Pearson's chi-square test

DISCUSSION

The present study found a significant decrease during the post-implementation period in the number of knee-related diagnostic claims per 1,000 insured persons in the intervention region compared to the control region. A similar decrease in the number of requested diagnostic imaging procedures for other joints was found. This decrease indicates that the implementation of the pathway went beyond awareness of requesting hip- and knee-related diagnostic imaging by GPs and positively influenced the GPs when it came to requesting diagnostic imaging in general. No differences in the number of hip-related diagnostic imaging procedures was found between the intervention and control region during the post-implementation period. Additionally, the pathway seems to have less effect on the referral behaviour of GPs. Regarding to the claims data, a significant difference in the number of initial orthopaedic consultations in the intervention region during the post-implementation period compared to the control region was not found. Furthermore, the random sample of the patient records showed that not all GPs seem to conform to the pathway since almost half of the patients were not referred to orthopaedic care according to the pathway. Besides this, patients with a suspicion of hip or knee OA referred to orthopaedic care according to the pathway had the same probability to undergo arthroplasty than patients referred not according to the pathway. This seems to indicate that the quality of referring did not improve through the implementation of the pathway.

The results of the present study regarding the absence of a significant decrease in the number of hip related diagnostic imaging are in line with the study of Linsell et al. [45]. in which GPs were more likely to request an x-ray for older people with hip pain than for older people with knee pain. A possible explanation for this could be the fact that hip complaints are more complex for GPs to manage. Literature shows that pain from the hip is difficult to define and that it is difficult to determine the exact source of pain [46, 47], that hip OA patients have more advanced complaints and that triggers for symptomatic presentation in hip OA are less understood [48]. When GPs experience difficulties in diagnosing hip related complaints, requesting diagnostic imaging can be a strategy to deal with these uncertainties [49]. Therefore, improving GPs skills to set the diagnosis OA of the hip could be the focus in future educational meetings. In addition, during the expert meetings, GPs revealed that it could be difficult to convince patients that diagnostic imaging is not always necessary to diagnose OA. Previous studies [50-52] have found that GPs' perception of patient pressure influences the non-adherence to guidelines concerning indications for

diagnostic imaging, like an X-ray or MRI. Moreover, Baker et al. [51] found that GPs believed that denying an X-ray could adversely affect the doctor–patient relationship. Although patient communication was part of the educational meetings, further improving GPs' patient-centered communication skills can be useful, since these skills are associated with fewer diagnostic testing expenditures [53].

An explanation for the lack of effect found in the present study regarding the referral behaviour of GPs may be the fact that the practical application of the pathway is not optimal since not all patients were referred to orthopaedic care according to the pathway. This could explain why no decreasing effect was found in the number of initial orthopaedic consultation claims. According to Rogers [54], when implementing an innovation, a part of the target group is sceptical and will offer resistance to change behaviour. Therefore, gaining insight into the application of the pathway by GPs may provide valuable information about the non-users. These insights can be used to evaluate barriers for application and to tailor interventions in order to stimulate the practical application [55]. Another explanation for the lack of effect could be the worldwide consistent increase in the incidence of joint arthroplasty [56]. This also explains the similar increase found in the control region. Additionally, during the expert meetings it emerged that patients can have a strong preference for a referral to orthopaedic care. Therefore, a referral sometimes is the only way to let patients accept that surgical treatment might not be beneficial. Literature confirms that patients' preferences and GPs' perception of patient pressure indeed influence the GP referral behaviour [52, 57]. This supports the evidence that guidelines are relatively ineffective when implemented on their own [58-60]. Again, improving patient-centered communication skills can be useful. Furthermore, increasing the consultation time per patient was mentioned during the expert meetings as an important criterion to apply these skills properly. Literature shows that longer consultations are associated with greater patient enablement [61], higher patient-centeredness [62], and a higher degree of offering lifestyle advice and preventive activities [63]. However, evidence about the influence of consultation length on the number of referrals and patient satisfaction is lacking [64]. In addition, exploring other interventions focussing on referring more appropriately to specialised medical care can be beneficial to reduce the inefficient use of limited resources [65-67]. Examples of such interventions are peer-reviewing referrals within a general practice before sending them to specialised medical care, enabling GPs to obtain the advice of medical specialists, periodic visits by medical specialists to GP practices, and shifts to outpatient clinics in which orthopaedic surgeons or other health-care

professionals with a special interest in musculoskeletal problems (for example GPs, nurse practitioners, or physician assistants) provide care in a community setting [67]. These alternatives appear promising with respect to reducing unnecessary referrals to specialised medical care but require further investigation into the effects on quality of care, patients' experiences, and cost of care [67].

Another point of attention mentioned during the expert meetings was the quality of physical therapy. Although the educational meetings and the distribution of the guidelines also aimed at physiotherapists, the focus of this study was on the impact of the implementation of the pathway on GPs behaviour. However, if physiotherapist do not use conservative treatment optimally and patients therefore do not experience improvements, GPs may feel forced to refer patients to orthopaedic care. Therefore, the practical implications of the pathway for physiotherapists and possibly other healthcare professionals (such as dieticians and psychologists) should be addressed in future research.

Based on the results, it is difficult to indicate which intervention (educational meetings, distribution of the guidelines, or reminders) contributed most to the decrease in the number of requested diagnostic imaging procedures related to the knee and other joints since this study did not examined the effect of the different interventions separately. However, in a systematic review conducted by French et al. [68] reminders were mentioned as potentially effective to change health professional behaviour and improve the use of diagnostic imaging. In the same review, educational meetings were not shown to be effective for changing imaging ordering behaviour. Furthermore, Hollingworth et al. [69] found no evidence about the effect of distributing clinical guidelines on changing GPs imaging behaviour related to patients with lumbar spine complaints. In addition, according to the literature [70], the educational meetings organised by members of the expert group have potential to impact on referral rates. More research is needed to learn about the effects of the various interventions within the context of this pathway. This information is needed to further optimise the implementation of the pathway and to achieve a further increase in the appropriate use of diagnostic imaging and possibly achieve a decrease in the number of referrals to orthopaedic care.

LIMITATIONS

In the present study, annual claims data were used. To ensure anonymity, only aggregated data (number of claims per year) were available and no analysis at an individual level could be made. Furthermore, claims data from only one health insurance company were used, which limits the ability to generalise the results of the study to a wider population (external validity) [71]. Since data from the health insurance company with the largest market share in the region were used, problems of selection bias are limited.

In addition, data on the exact extent to which the pathway was applied at GP level in the intervention region were missing. Although all GPs were informed about the pathway, information about how many GPs requested diagnostic imaging procedures and referred patients to orthopaedic care according to the guidelines was lacking. This makes it difficult to attribute the implementation results of the pathway and to consider if there is more room for improvement. Therefore, more research is needed to measure the 'real' effect of the pathway in the future.

Furthermore, the present study focused merely on the effects of the implementation of the pathway on GPs behaviour. However, healthcare professionals like physiotherapists, dieticians and psychologist are also involved in the conservative treatment of patients with hip and knee OA. Therefore, research on the effects of the pathway across the entire width of care for hip and knee OA is necessary in order to improve the effect of the pathway.

Finally, it is important to focus not only on the number of requested diagnostic imaging procedures and referrals to orthopaedic care, but also on the effect of the pathway on patient satisfaction, quality and costs of care [72]. Therefore, more extensive research in patients, for example through the use of questionnaires, is needed [73].

CONCLUSION

The introduction of a pathway aiming to reduce GP diagnostic imaging requests and GP referrals to orthopaedic surgeons for hip and knee OA, had mixed effects. Results showed a decrease in the number of diagnostic imaging requests for knee and other joint related OA, but no impact was found on those for hip OA. In parallel, referrals to orthopaedic care increased during the post-implementation period, both for hip and knee OA related referrals. Future research is needed to identify the specific role

of the interventions in their effectiveness in improving the diagnostic behaviour of GPs, particularly related to diagnostic imaging procedures of the hip. In addition, further research on the referral behaviour of GPs is necessary, which should focus on possible other interventions and the entire width of care for hip, and knee OA in order to improve the effect of the pathway.

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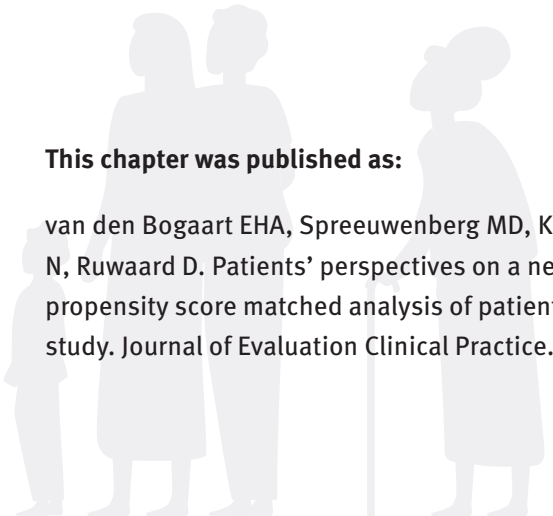
CHAPTER 5

Patients' perspectives on a new delivery model in primary care

A propensity score matched analysis of patient-reported outcomes in a Dutch cohort study



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ABSTRACT

Rationale, aims and objective: Primary Care Plus (PC+) focuses on the substitution of hospital-based medical care to the primary care setting without moving hospital facilities. The aim of this study was to examine whether population health and experience of care in PC+ could be maintained. Therefore, health-related quality of life (HRQoL) and experienced quality of care from a patient perspective were compared between patients referred to PC+ and to hospital-based outpatient care (HBOC).

Methods: This cohort study included patients from a Dutch region, visiting PC+ or HBOC between December 2014 and April 2018. With patient questionnaires (T0, T1 and T2), the HRQoL and experience of care were measured. One-to-two nearest neighbor caliper propensity score matching (PSM) was used to control for potential selection bias. Outcomes were compared using marginal linear models and Pearson chi-square tests.

Results: 1,113 PC+ patients were matched to 606 HBOC patients with well-balanced baseline characteristics (SMDs ‹ 0.1). Regarding HRQoL outcomes, no significant interaction terms between time and group were found (P › 0.05), indicating no difference in HRQoL development between the groups over time. Regarding experienced quality of care, no differences were found between PC+ and HBOC patients. Only travel time was significantly shorter in the HBOC group ($P \leq 0.001$).

Conclusion: Results show equal effects on HRQoL outcomes over time between the groups. Regarding experienced quality of care, only differences in travel time were found. Taken as a whole, population health and quality of care were maintained with PC+ and future research should focus more on cost-related outcomes.

INTRODUCTION

In 1978, the Declaration of Alma Ata identified primary health care as the key to achieving the goal of delivering better health care for all [1]. Forty years later, the Global Conference on Primary Health Care came with a renewed declaration, in which the importance of focusing on primary health care was emphasised again [2]. This new declaration states that, a focus on primary health care is still critical due to growing possibilities of technology, an ageing population and an increasing number of people suffering from multimorbidity [3-5]. These developments lead to rapidly increasing health care costs in developed countries [6]. According to the OECD, public expenditure on health- and long-term care will increase to 9% of Gross Domestic Product in 2030 and even to 14% by 2060 in OECD countries. Therefore, the future sustainability of health care systems is at stake. Governments are challenged to continue providing accessible, equitable and affordable health care of adequate quality. In order to do so, policymakers are forced to redesign health care delivery models [4].

As primary care functions as the door to the whole health care system, strengthening primary care is an important policy instrument in redesigning health care [5]. An example is to shift hospital-based medical specialists to the primary care setting without moving the hospital facilities [7-10]. This shift is a form of substitution, defined as: 'the continual regrouping of resources across and within care settings, to exploit the best and least costly solutions in the face of changing needs and demands' [11]. In 2013, regional collaboration initiatives in the Netherlands, focusing on substitution, were established to achieve the Triple Aim by improving the experience of care and the health of the population, and reducing the per capita costs [12, 13] Primary Care Plus (PC+) is one of these initiatives [14-16].

With the Triple Aim framework, Berwick et al [13] encourages health care organisations to reduce the cost of care, while at the same time increase the health of the population and the quality of care. In a study by Quanjel et al. [17], a PC+ intervention for patients with cardiology-related complaints was evaluated based on the principles of the Triple Aim. In this PC+ setting, cardiologists provided consultations in the presence of similar diagnostic tools as in the hospital. The results showed that besides cost reduction, the health of the population and the quality of care did not decrease compared to care-as-usual. However, the present study focusses on a PC+ intervention in which hospital facilities are not available and therefore, medical specialists are only able to use their own expertise and

experience. This forces them to use a generalist approach to analyse a patient's medical complaint [18].

This study aims to evaluate whether the PC+ initiative (without the availability of the hospital facilities), is also able to increase the health of the population and the quality of care. Therefore, the health-related quality of life (HRQoL) and the experienced quality of care from the patient's perspective are compared between patients referred to PC+ and patients referred directly to hospital-based outpatient care (HBOC).

METHODS

Study design

This cohort study compared patient-reported HRQoL and the experienced quality of care between patients referred to PC+ and patients referred to HBOC using propensity score matching (PSM). The reporting of this study follows the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines [19].

The study is approved by the Medical Research and Ethics Committee of the Maastricht University Medical Centre (METC 14-4-136). Informed consent was obtained from all individual participants included in the study.

Setting and intervention

In the Maastricht–Heuvelland region, located in the southern Netherlands, the primary care organisation Care in Development (in Dutch 'Zorg in ontwikkeling'), Maastricht University Medical Centre + (Maastricht UMC+), health insurance company VGZ and patient representative foundation 'Burgerkracht Limburg' collaborate. In 2014, these organisations developed the PC+ intervention to substitute hospital-based specialised care with primary care whereby GPs remain responsible for the patient. With two PC+ centres operating according to the same method, GPs within the region are able to refer non-acute and low-complex patients to a medical specialist in a neutral primary care setting. Based on the PC+ patients' profiles (listing relevant medical complaints for PC+), GPs' clinical expertise and shared decision-making, GPs decide whether to refer a patient to PC+. In PC+, the medical specialist examines and/or treats the patient during a maximum of two consultations. Following PC+, the medical specialist refers the patient back to the GP with treatment advice, or, if necessary, refers the patient to HBOC for further diagnosis and/or treatment. Involved specialists are employed in the Maastricht UMC+ and perform

PC+ consultations on a regular basis (weekly or biweekly). Like the Maastricht UMC+, the PC+ centres are both located in the city of Maastricht.

Besides the assumed benefits of PC+ being more informal and located closer to patients' homes, patients are exempt from paying a mandatory deductible for a consultation. In the Netherlands, GP consultations are fully covered by health insurance but for consulting a medical specialist, a yearly mandatory deductible is levied (€360 in 2014 and €385 since 2016) [20]. This mandatory deductible is determined by the government [21]. Patients have to pay this deductible themselves before the health insurance company pays for specialised medical care.

Study population

In 2016, The Maastricht–Heuvelland region consisted of 55 GP practices caring for a population of about 170,000 people [22]. Patients eligible for inclusion were adult patients (≥ 18 years) from the Maastricht–Heuvelland region visiting PC+ or HBOC between December 2014 and April 2018, with a referral to one of the medical specialties present in PC+ during the study period: dermatology, gynaecology, otolaryngology, internal medicine (including gastroenterology), neurology, ophthalmology, orthopaedics, rheumatology and urology. This study is part of a larger study, which requires 1,830 patients per group (3,660 patients in total) [16].

Data collection

After referral to PC+ or HBOC by the GP, all eligible patients were recruited by the Transmural Interactive Patient Platform (TIPP) for participation. TIPP plans and registers referrals to medical specialists in either PC+ or HBOC. TIPP informed patients about the study, and if interested, patients' contact details were sent to the research team. The research team then sent an information letter, informed consent and the first questionnaire (T0) to the patient by post or email. Patients were asked to return the informed consent and the questionnaire before the first consultation with the medical specialist. After the first consultation, a second questionnaire was sent within one week (T1) and a third questionnaire after three months (T2). The inclusion of patients started in December 2014 and continued until April 2018.

Outcome measures

BASELINE CHARACTERISTICS

Baseline characteristics were collected during T0, including age in years, gender, native country and level of education (low vs medium vs high) (Figure 1). Collected risk

factors included body mass index (BMI) calculated from reported height and weight, cigarette smoking (current vs former vs never) and alcohol use (yes vs no).

HEALTH-RELATED QUALITY OF LIFE

To measure generic HRQoL, the EuroQol five-dimensional questionnaire with five levels (EQ-5D-5L), including the EuroQol Visual Analogue Scale (EQ-VAS) and the Short-Form Health Survey version 2 (SF-12v2) were used. Patients' perceptions of a change in their health status was evaluated with the Patient Global Impression of Change (PGIC) seven-item response scale.

The EQ-5D-5L is a measure consisting of five questions on mobility, self-care, pain/discomfort, usual activities and anxiety/depression with five response levels [23]. A health state index score, ranging from -0.446–1 (worst to best imaginable health status), was calculated from individual health profiles using the Dutch utility tariff [24]. The included EQ-VAS is a 0–100 scale where respondents indicate their overall health. Both the EQ-5D-5L and the EQ-VAS were measured at T0, T1 and T2. The minimal clinically important change in EQ-5D-5L is 0.04 [25].

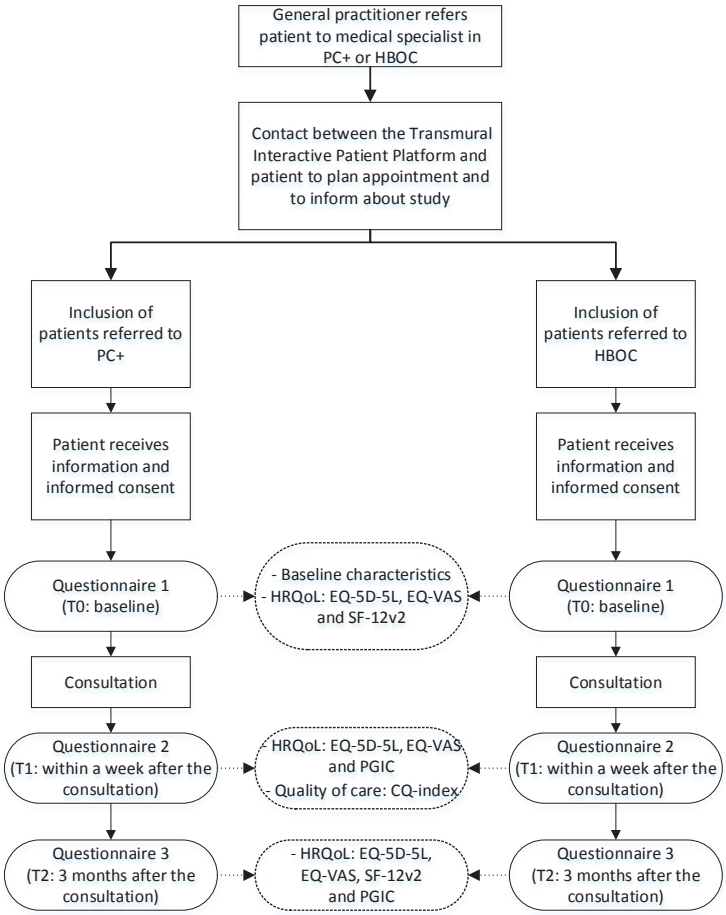
The SF-12v2 consists of 12 questions measuring the health status by means of two summary scores; a physical component summary (PCS) and a mental component summary (MCS)[26]. PCS and MCS scores range from 0 (lowest level of health) to 100 (highest level of health) and were obtained using the instrument developers' standard scoring algorithm [26]. The SF-12v2 was measured at T0 and T2. The minimal clinically important change for both PCS and MCS scores ranges between 3–5 points [27].

With the PGIC scale, patients were able to indicate to what extent their health problem had changed after they consulted the medical specialist, ranging from 1 (very much improved) to 7 (very much worse) [28]. The PGIC scale was conducted at T1 and T2.

EXPERIENCED QUALITY OF CARE

An influential and often used framework to measure quality of care is that of the Institute of Medicine, stating care must be safe, effective, patient-centered, timely, efficient and equitable [29]. Patient centeredness and timeliness are explicitly included in the Consumer Quality (CQ) index. The CQ-index is a standardised method for measuring experiences of patients with health care [30]. In this study, 21 items derived from the Dutch CQ-index general practice [31]. and hospital outpatient care [32]. were used. Items can be divided into five domains: timeliness (3 items);

treatment by the medical specialist (6 items); information provision and communication by the medical specialist (4 items); communication and collaboration between the medical specialist and GP (4 items); and the overall assessment of quality of care (4 items). Most item scores ranged from 1–4. However, travel time was measured in minutes on a continuous scale. Furthermore, the medical specialist and the outpatient clinic visited were graded on a 0–10 scale. The CQ-index was measured at T1.



Note: PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; HRQoL = Health Related Quality of Life; EQ-5D-5L = EuroQol five-dimensional questionnaire with five levels; EQ-VAS = EuroQol Visual Analogue Scale; SF-12 = Short-Form 12 item (version 2) Health Survey; PGIC = Patient Global Impression of Change; CQ-index = Consumer Quality index

Figure 1 Flow Chart of Participating Patients Flow and Questionnaire Measurements

Statistical methods

NON-RESPONSE

Non-response analysis was performed by comparing respondents with non-respondents at baseline by patient age, gender and the medical specialty referred to.

PROPENSITY SCORE MATCHING

Since in this cohort study patients were not-randomly allocated to treatment, patients being referred to PC+ were expected to differ on covariates to those referred to HBOC [33, 34]. To correct for this potential selection bias, which may affect the estimates of the treatment effect, PSM was used [35]. First, the propensity score (PS) was estimated using logistic regression, which predicts the likelihood of a referral to PC+ or HBOC based on the baseline characteristics described earlier. By matching patients in the intervention and control group based on the PS, the groups will be more balanced on the observed baseline characteristics, which enables to obtain less biased estimates of treatment effects. In this study, one-to-two nearest neighbour caliper matching without replacement was used, with a caliper of 0.1 [36]. One-to-two matching was used to keep a larger sample size since the HBOC group was small [37]. Baseline characteristics before and after matching were compared with *P*-values and standardised mean differences (SMDs). SMDs of ‹ 0.1 and *P-value*s of › 0.05 indicate minor differences in the mean of a covariate between the two groups and were used to assess the success of matching [38].

COMPARING STUDY GROUPS

The overlap in the distribution of the PS and the balance of baseline variables before and after matching between the PC+ and HBOC groups were described.

Marginal linear models with an unstructured error covariance structure were applied to analyse the mean change in HRQoL outcome measures. Estimates, standard errors (SEs), 95% confidence intervals (CIs) and *P*-values were reported. *P*-values ‹ 0.05 were considered as significant. This method takes into account incomplete follow-up data without any imputation of missing values, and provides valid estimates of treatment effects under the assumption that such data are missing at random [39].

Patients' experiences of care related items were dichotomised before analysing and summarised as 'satisfied' versus 'unsatisfied' or 'yes' versus 'no'. Hereafter, they were analysed using Pearson chi-square tests; counts, percentages and *P*-values

were reported. Additionally, independent t-tests were used to analyse continuous items; 95% CIs and *P*-values were reported. To correct for multiple testing (Type 1 error) the Bonferroni correction was used, whereby the *P*-value of 0.05 was divided by the number of tests [40]. Furthermore, analyses were applied without imputation of missing data and items with a high non-response (more than 10% missing values) were excluded.

Before taking into account the influence of the PS on the HRQoL and experienced quality of care outcome measures using PSM, the uncorrected effect of 'study group' was analysed, with 'study group' (PC+ vs HBOC) as the only independent variable [41].

R Studio was used for statistical analyses (R Studio, Boston, MA).

SUBGROUP ANALYSES

Baseline characteristics, HRQoL and experience of care outcomes before and after PSM were compared between PC+ and HBOC patients separately for the nine different medical specialties using the same analyses as described above.

SENSITIVITY ANALYSES

Sensitivity analyses were undertaken to assess the robustness of the results [42]. Analyses were repeated using a one-to-one nearest neighbour caliper matching without replacement with a caliper of 0.1.

RESULTS

Study participants and responders' characteristics

Figure 2 presents a flow chart detailing the inclusion and exclusion of patients. Contact details of 5,535 patients were sent to the research team (n = 3,890 (70.3%) PC+ group and n = 1,645 (29.7%) HBOC group). In total, 2,898 patients responded to the informed consent and/or first questionnaire (n = 2,120 (54.5%) PC+ group and n = 778 (47.3%) HBOC group). However, the first questionnaire (T0) was not completed by all patients. The first questionnaire was completed by 2,076 PC+ patients (53.4%) and 761 HBOC patients (46.3%). Because of missing both follow-up questionnaires (T1 and T2), 313 (15.1%) PC+ patients and 118 (15.5%) HBOC patients were excluded. As a result, 1,763 PC+ patients and 643 HBOC patients were eligible for matching (total N = 2,406).

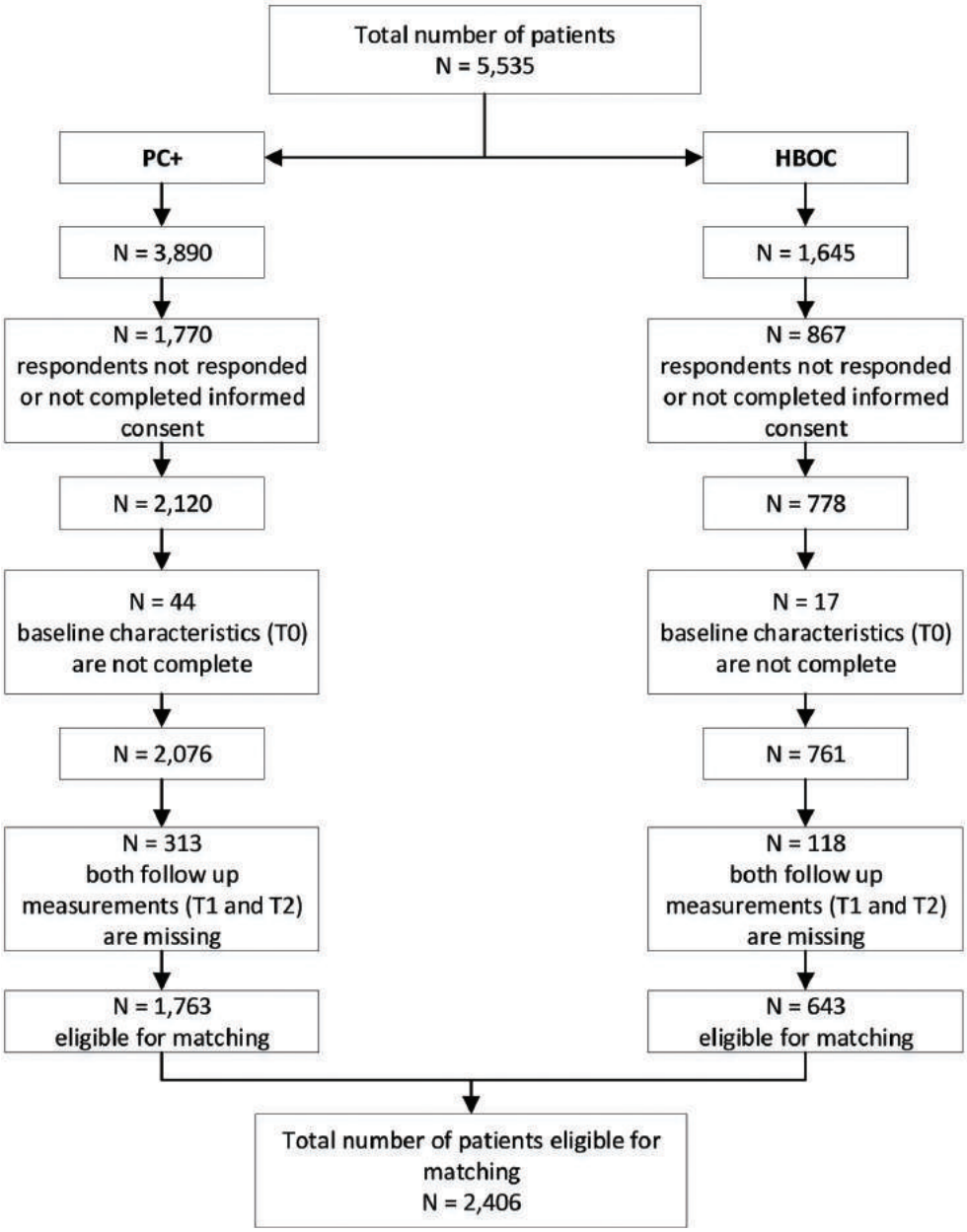
The characteristics of the 2,898 responders and 2,637 non-responders are attached in the supplementary Table S1. Responders in the PC+ and HBOC group were significantly older compared to non-responders. Regarding the medical specialty referred to; there was a significant difference in the distribution between responders and non-responders in the HBOC group, with proportionally more responders referred to ophthalmology, otolaryngology and dermatology.

Inspection for PS overlap before and after matching

Before PSM, the PS for the PC+ group ranged between 0.08 and 0.73; for the HBOC group, the PS ranged between 0.09 and 0.78 (see Figure 3). After PSM, the PS for the PC+ group ranged between 0.10 and 0.73; for the HBOC group, the PS ranged between 0.10 and 0.74.

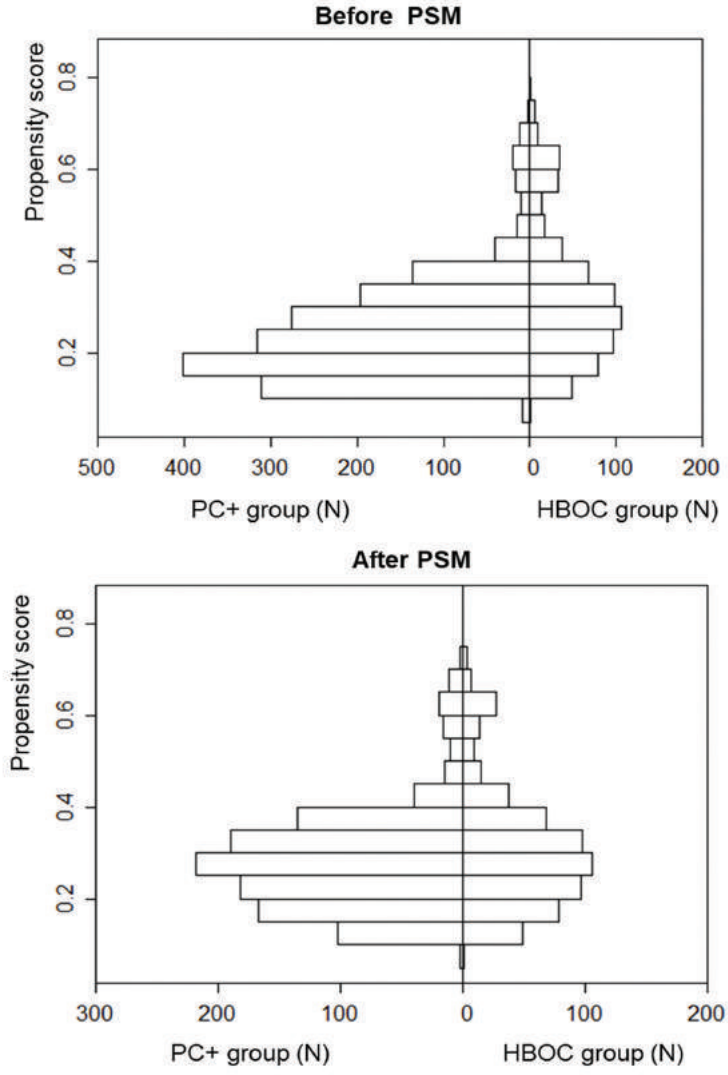
Baseline characteristics

Prior to PSM, PC+ patients were younger and had a better HRQoL at baseline (Table 1). Furthermore, respectively more PC+ patients were referred to dermatology and rheumatology, and less to internal medicine, neurology, orthopaedics and urology. After PSM, with 1,113 PC+ patients matched to 606 HBOC patients, these characteristics were well balanced with a SMD < 0.1, except for the percentage of patients referred to internal medicine (SMD = 0.145).



Note: PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

Figure 2 Flow Chart of Study Inclusion



Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

Figure 3 Overlap of the Propensity Score in the Two Study Groups

Table 1 Baseline characteristics before and after propensity score matching

	Before PSM	
	PC+	
N	**1763**	
Age (mean, SD)	55.95	15.68
Gender (male) (%, SD)	39%	0.49
Native country (Netherlands) (%, SD)	97%	0.18
Educational level		
Low (%, SD)	19%	0.39
Medium (%, SD)	47%	0.50
High (%, SD)	34%	0.48
EQ-5D-5L (mean, SD)	0.81	0.17
EQ-VAS (mean, SD)	75.53	16.32
SF12 PCS (mean, SD)	47.44	9.33
SF12 MCS (mean, SD)	51.22	9.35
BMI (mean, SD)	26.16	4.45
Smoking behaviour		
Smoker (%, SD)	16%	0.36
Former smoker (%, SD)	42%	0.49
Non-smoker (%, SD)	42%	0.49
Alcohol user (%, SD)	62%	0.48
Medical specialty referred to		
Dermatology (%, SD)	32%	0.47
Gynaecology (%, SD)	5%	0.22
Internal medicine (%, SD)	2%	0.15
Otolaryngology (%, SD)	17%	0.37
Neurology (%, SD)	7%	0.26
Ophthalmology (%, SD)	9%	0.28
Orthopaedics (%, SD)	19%	0.39
Rheumatology (%, SD)	7%	0.25
Urology (%, SD)	1%	0.12

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	HBOC		P-value	SMD
	643			
	57.63	15.23	0.019*	0.109
	41%	0.49	0.557	0.027
	96%	0.20	0.393	0.038
	22%	0.42	0.063	0.084
	46%	0.50	0.593	0.025
	32%	0.47	0.317	0.046
	0.79	0.17	0.005**	0.129
	73.08	16.31	0.001**	0.150
	45.39	10.04	≤0.001***	0.211
	50.11	9.34	0.010**	0.119
	26.44	4.84	0.178	0.061
	17%	0.38	0.443	0.035
	42%	0.49	0.771	0.013
	41%	0.49	0.781	0.013
	60%	0.49	0.198	0.059
	16%	0.37	≤0.001***	0.371
	7%	0.25	0.260	0.051
	9%	0.29	‹0.001***	0.307
	13%	0.34	0.046	0.094
	12%	0.33	≤0.001***	0.159
	9%	0.28	0.821	0.010
	24%	0.43	0.009**	0.119
	4%	0.19	0.005**	0.138
	6%	0.23	≤0.001***	0.239

Table 1 Continued

	After PSM	
	PC+	
N	**1113**	
Age (mean, S D)	57.88	14.60
Gender (male) (%, SD)	39%	0.49
Native country (Netherlands) (%, SD)	96%	0.20
Educational level		
Low (%, SD)	21%	0.41
Medium (%, SD)	46%	0.50
High (%, SD)	33%	0.47
EQ-5D-5L (mean, SD)	0.79	0.18
EQ-VAS (mean, SD)	73.34	16.75
SF12 PCS (mean, SD)	45.59	9.55
SF12 MCS (mean, SD)	50.77	9.46
BMI (mean, SD)	26.53	4.57
Smoking behaviour		
Smoker (%, SD)	16%	0.37
Former smoker (%, SD)	42%	0.49
Non-smoker (%, SD)	42%	0.49
Alcohol user (%, SD)	60%	0.49
Medical specialty referred to		
Dermatology (%, SD)	19%	0.39
Gynaecology (%, SD)	7%	0.25
Internal medicine (%, SD)	4%	0.19
Otolaryngology (%, SD)	16%	0.36
Neurology (%, SD)	11%	0.32
Ophthalmology (%, SD)	11%	0.31
Orthopaedics (%, SD)	26%	0.44
Rheumatology (%, SD)	4%	0.20
Urology (%, SD)	2%	0.15

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	HBOC		P-value	SMD
	606			
	57.85	15.13	0.960	0.003
	41%	0.49	0.498	0.034
	96%	0.20	0.876	0.008
	22%	0.42	0.511	0.033
	46%	0.50	0.960	0.003
	32%	0.47	0.531	0.032
	0.79	0.17	0.896	0.007
	73.10	16.37	0.767	0.015
	45.37	10.05	0.645	0.023
	50.28	9.34	0.302	0.052
	26.54	4.82	0.961	0.002
	17%	0.38	0.591	0.027
	42%	0.49	0.948	0.003
	41%	0.49	0.736	0.017
	59%	0.49	0.810	0.012
	17%	0.38	0.430	0.040
	7%	0.25	0.992	0.001
	7%	0.25	0.003**	0.145
	14%	0.35	0.454	0.038
	12%	0.33	0.516	0.033
	9%	0.29	0.264	0.057
	26%	0.44	0.706	0.019
	4%	0.20	0.728	0.018
	4%	0.19	0.070	0.088

Outcome analysis

HEALTH-RELATED QUALITY OF CARE

Before PSM, the EQ-5D-5L baseline score was significantly lower in the HBOC group ($P < 0.01$) (Table 2). After PSM, the difference at baseline between PC+ and HBOC patients was no longer significant ($P > 0.05$). Furthermore, the EQ-5D-5L scores significantly increased over time (T1 and T2) compared to the baseline score before and after PSM ($P < 0.01$ or $P < 0.001$). Finally, after PSM, the interaction terms between time and group were no longer significant, indicating no difference in the development of EQ-5D-5L scores between the groups over time ($P > 0.05$).

Regarding EQ-VAS outcomes, before PSM, the baseline score was significantly lower in the HBOC group compared to the PC+ group ($P < 0.01$). After PSM, the difference at baseline was no longer significant ($P > 0.05$). Furthermore, EQ-VAS scores significantly increased at T1 compared to the baseline score before and after PSM ($P < 0.01$). However, no significant interaction terms between time and group were found before and after PSM, indicating no difference in the development of EQ-VAS scores between the groups over time ($P > 0.05$).

Regarding SF12v2 scores, before PSM, the PCS and MCS baseline scores were significantly lower in the HBOC group ($P < 0.001$ and $P < 0.01$, respectively). After PSM, the differences at baseline were no longer significant ($P > 0.05$). Furthermore, before and after PSM, the PCS score at T2 was significantly higher compared to the baseline score. However, for both PCS and MCS, no significant interaction terms were found before and after PSM, indicating no difference in the development of the PCS and MCS scores between the groups over time ($P > 0.05$).

Finally, the PGIC score at T1 did not differ between the PC+ and HBOC groups ($P > 0.05$). At T2, the PGIC score was significantly lower compared to the score at T1, both before and after PSM ($P < 0.05$ and $P < 0.001$, respectively). However, no significant interaction terms between time and group were found before and after PSM, indicating no difference in the development of the PGIC score between the groups over time ($P > 0.05$).

Figures for the HRQoL outcomes before and after PSM are attached in the supplementary Figure S1.

In total, 2,365 patients completed the second questionnaire (T1) including the 21 items of the CQ-index. (n = 1,741 PC+ group and n = 624 HBOC group). After PSM, 1,681 patients were included in the analysis (n = 1,094 PC+ group and n = 587 HBOC group).

One item in the domain of 'communication and collaboration between the GP and medical specialist' was excluded from analysis because of high non-response before (13.5%) and after (13.1%) PSM. Although, only 1,230 patients before and 900 patients after PSM completed the item 'shared decision-making', this item was not excluded since a high number of patients answered 'not applicable'. This was the only item in the questionnaire with this answering option. Including the option 'not applicable', 2,320 patients (98.1%) completed this item before PSM and 1,659 patients (98.7%) after PSM.

Before PSM, PC+ patients significantly more often had a waiting time in the waiting room of less than 30 minutes ($P \leq 0.001$) and they gave significantly higher grades to the medical specialist and the PC+ location the visited ($P = 0.007$ and $P \leq 0.001$, respectively) (Table 3). However, after PSM, these differences were no longer significant ($P = 0.011$, $P = 0.199$ and $P = 0.354$, respectively). Furthermore, before PSM, the travel time to the PC+ or HBOC location was significantly shorter in the PC+ group ($P \leq 0.001$). However, after PSM, the travel time was significantly shorter in the HBOC group ($P \leq 0.001$).

Table 2 Health-related quality of life outcomes before and after propensity score matching

	Before PSM		
EQ-5D-5L	**Estimate**	**SE**	**95% CI**
Intercept	0.82***	0.00	0.81 - 0.82
Study group [A]	−0.02**	0.01	−0.04 - −0.01
Time T1	0.01***	0.00	0.01 - 0.02
Time T2	0.02***	0.00	0.01 - 0.02
Time T1 x study group	−0.01*	0.01	−0.02 - 0.00
Time T2 x study group	−0.01*	0.01	−0.03 - −0.00
EQ-VAS	**Estimate**	**SE**	**95% CI**
Intercept	75.53***	0.39	74.77 - 76.29
Study group [A]	−2.45**	0.75	−3.92 - −0.97
Time T1	0.92**	0.31	0.32 - 1.53
Time T2	1.10	0.64	−0.16 - 2.35
Time T1 x study group	−0.41	0.60	−1.58 - 0.77
Time T2 x study group	−0.16	1.25	−2.60 - 2.28
SF-12 PCS	**Estimate**	**SE**	**95% CI**
Intercept	47.44***	0.23	46.99 - 47.88
Study group [A]	−2.05***	0.44	−2.91 - −1.19
Time T2	0.57***	0.17	0.24 - 0.90
Time T2 x study group	0.11	0.33	−0.53 - 0.75
SF-12 MCS	**Estimate**	**SE**	**95% CI**
Intercept	51.22***	0.22	50.78 - 51.66
Study group [A]	−1.11**	0.43	−1.95 - −0.27
Time T2	0.03	0.20	−0.36 - 0.42
Time T2 x study group	−0.32	0.39	−1.08 - 0.44
PGIC B	**Estimate**	**SE**	**95% CI**
Intercept	4.57***	0.73	3.14 - 6.01
Study group A	−1.10	1.41	−3.88 - 1.67
Time T2	−1.58*	0.73	−3.02 - −0.15
Time T2 x study group	1.11	1.42	−1.67 - 3.89

Note: PSM = propensity score matching; SE = standard error; CI = confidence interval

** P < 0.05; ** P < 0.01; *** P < 0.001; [A] Group was coded as 1 = Hospital Based Outpatient Care (HBOC) group and 0 = Primary Care Plus (PC+) group; [B] PGIC was measured at T1 and T2, not at baseline*

After PSM		
Estimate	**SE**	**95% CI**
0.79***	0.01	0.78 - 0.80
−0.00.	0.01	−0.02 - 0.02
0.01**	0.00	0.00 - 0.02
0.02***	0.00	0.01 - 0.02
−0.01	0.01	−0.02 - 0.00
−0.01	0.01	−0.03 - 0.00
Estimate	**SE**	**95% CI**
73.34***	0.50	72.37 - 74.32
−0.25	0.84	−1.89 - 1.40
1.11**	0.42	0.29 - 1.92
1.59	0.91	−0.19 - 3.38
−0.66	0.70	−2.03 - 0.72
−0.62	1.54	−3.64 - 2.40
Estimate	**SE**	**95% CI**
45.59***	0.29	45.02 - 46.17
−0.23	0.49	−1.19 - 0.74
1.02***	0.22	0.59 - 1.45
−0.36	0.37	−1.08 - 0.37
Estimate	**SE**	**95% CI**
50.77***	0.28	50.21 - 51.32
−0.49	0.48	−1.42 - 0.44
0.21	0.25	−0.28 - 0.71
−0.65	0.43	−1.49 - 0.20
Estimate	**SE**	**95% CI**
3.45***	0.03	3.40 - 3.51
0.03	0.05	−0.06 - 0.13
−0.35***	0.04	−0.43 −0.27
−0.14*.	0.07	−0.28 - 0.01

Table 3 Comparison of patient-experienced quality of care outcomes before and after propensity score matching

	Before PSM		
	PC+	HBOC	
N	1741	624	
Quality of care domains	Satisfied/Yes n (%)	Satisfied/Yes n (%)	P-value
Timeliness (1)			
Waiting time for appointment	89.2(1527)	86.0(533)	0.032
Waiting time in waiting room ‹30 minutes	93.5 (1605)	88.5 (546)	≤0.001*
Treatment by the medical specialist			
Complaint was taken seriously	97.5(1672)	97.4(601)	0.845
Specialist listened carefully	97.3(1667)	97.4(601)	0.845
Specialist took enough time	98.0(1679)	98.7(608)	0.240
Treated with respect	98.8(1692)	98.5(607)	0.574
Competence of the specialist	98.4(1673)	98.0(601)	0.543
Overall help of the specialist	94.2(1612)	93.5(575)	0.553
Information provision and communication by the medical specialist			
Information about different treatment options	92.6(1581)	90.7(555)	0.140
Understandable explanation	97.1(1663)	96.1(592)	0.236
Opportunity to ask questions	97.4(1666)	96.4(594)	0.231
Shared decision-making	88.4(892)	87.3(338)	0.582
Communication and collaboration between the GP and medical specialist			
Matching recommendations between GP and specialist	80.3(1357)	82.1(501)	0.324
Awareness of the medical specialist about the complaint	89.4(1519)	89.1(547)	0.827
Collaboration and alignment between GP and specialist	85.8(1366)	81.6(482)	0.016

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; GP = general practitioner; SD = standard deviation

** P ‹ 0.0025 were considered as significant according to the Bonferroni correction; [A] A significant higher score on travel time means a longer travel time in minutes and is in this case an unfavourable result*

	After PSM		
	PC+	HBOC	
	1094	587	
	Satisfied/Yes n (%)	Satisfied/Yes n (%)	P-value
	89.9(972)	86.5(505)	0.034
	92.0(997)	88.1(513)	0.011
	97.0(1052)	97.3(566)	0.813
	96.9(1050)	97.3(566)	0.659
	97.9(1061)	98.6(573)	0.284
	98.4(1066)	98.5(572)	0.974
	98.5(1063)	98.1(567)	0.520
	94.0(1018)	93.3(541)	0.562
	92.2(998)	90.5(523)	0.220
	97.4(1055)	95.9(557)	0.084
	97.2(1055)	95.9(557)	0.256
	88.3(580)	87.0(320)	0.535
	79.1(846)	82.1(472)	0.153
	88.1(946)	89.5(518)	0.399
	85.2(859)	81.7(454)	0.066

Table 3 Continued

	Before PSM		
	PC+	**HBOC**	
N	**1741**	**624**	
Quality of care domains	**Satisfied/Yes n (%)**	**Satisfied/Yes n (%)**	**P-value**
Overall assessment of quality of care (1)			
Recommend medical specialist to family/friends	93.7(1598)	92.5(568)	0.298
Recommend PC+/HBOC to family/ friends	95.4(1625)	93.8(577)	0.119
	Mean (SD)	**Mean (SD)**	
Timeliness (2)			
Travel time (in minutes) [A]	15.6 (9.34)	19.2 (12.18)	≤0.001*
Overall assessment of quality of care (2)			
Grade specialist (0–10)	8.5 (1.15)	8.4 (1.22)	0.007
Grade PC+/HBOC (0–10)	8.5 (1.08)	8.3 (1.11)	≤0.001*

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; GP = general practitioner; SD = standard deviation

** P < 0.0025 were considered as significant according to the Bonferroni correction; [A] A significant higher score on travel time means a longer travel time in minutes and is in this case an unfavourable result*

	After PSM		
	PC+	**HBOC**	
	1094	**587**	
	Satisfied/Yesn (%)	**Satisfied/Yes n (%)**	**P-value**
	92.9(1002)	92.2(544)	0.506
	95.1(1024)	93.8(544)	0.268
	Mean (SD)	**Mean (SD)**	
	17.0 (10.75)	15.6 (9.10)	0.001*
	8.5 (1.15)	8.5 (1.20)	0.199
	8.4 (1.08)	8.5 (1.12)	0.354

Subgroup analyses

In the subgroup analyses, the baseline characteristics, HRQoL and experiences of care related outcomes before and after PSM were analysed per medical specialty. Regarding baseline characteristics, all medical specialties had two or more characteristics with a SMD › 0.1, indicating less balanced groups (see supplementary Table S3).

Regarding HRQoL outcomes, significant interactions between time and group after PSM were found for the medical specialties neurology, otolaryngology and internal medicine, indicating a positive effect for PC+ patients over time (see supplementary Table S3). Time effects were found for neurology on the EQ-5D-5L at T1 and on the EQ-VAS at T1 and T2, for otolaryngology on the SF12v2 MCS and the PGIC, and for internal medicine on the SF12v2 MCS. However, for dermatology, a negative effect was found on the SF12v2 PCS score, indicating that HBOC resulted in better outcomes on the physical component over time compared to PC+.

Regarding experienced quality of care outcomes measured on 20 items, after PSM PC+ scored higher on three items for dermatology and on one item for neurology (see supplementary Table S4). Furthermore, a significantly higher score on travel time (meaning a longer travel time) was found for HBOC patients referred to dermatology, otolaryngology and orthopaedics.

Sensitivity analyses

After one-to-one PSM, the PS for the PC+ group ranged between 0.09 and 0.73; for the HBOC group, the PS ranged between 0.10 and 0.74 (see supplementary Figure S2). In total, 609 PC+ patients were matched to 609 HBOC patients with well-balanced baseline characteristics (all SMD ‹ 0.1 and *P-value*s › 0.05; see supplementary Table S5). Regarding HRQoL outcome analysis after one-to-one PSM, the results were comparable to one-to-two PSM with no significant interaction terms between time and group (see supplementary Table S6). Regarding experienced quality of care after one-to-one PSM, most results were comparable to one-to-two PSM (see supplementary Table S7). However, the difference in travel time to the PC+ or HBOC location was no longer significant ($P = 0.212$).

DISCUSSION

In this study, PSM resulted in balanced groups with respect to measured baseline characteristics. Therefore, the effects of PC+ on the health of the population and patients' experiences of care could be compared to HBOC. The results showed that PC+ care for low-complex and non-acute patients delivered in a primary care setting without the presence of hospital facilities led to the maintenance of patients' experiences of HRQoL and quality of care.

These results are generally consistent to those of Quanjel et al. [17] who evaluated a PC+ intervention focusing on cardiologists providing consultations in a primary care setting. They concluded that PC+ results in equal effects on HRQoL outcomes over time and improved quality of care as experienced by patients compared to care-as-usual. Other studies including shifted HBOC also found high levels of patient satisfaction [43, 44].

This study showed positive results regarding patients' experiences of HRQoL in PC+. To measure HRQoL, generic instruments were used since they are applicable to all patients, regardless of the medical specialty referred to and regardless of the patient's condition. Therefore, comparison between different medical specialties and interventions is possible [45]. However, generic instruments are limited in detecting change over time (responsiveness) compared to disease- or condition-specific instruments [46]. Therefore, equal effect on HRQoL outcomes could be the result of the use of generic instruments to measure the HRQoL over time. In future research, using both generic and condition-specific instruments should be considered to increase responsiveness.

Furthermore, this study showed that patients were highly satisfied with the care delivered in PC+. This is a positive result, although it is recognised that patients remain reluctant to be critical about the care they receive [47]. This is based on patient desire to be grateful, as well as their recognition of the inevitable limitations of health care. However, patient satisfaction could be supplemented with clinical outcome measures focused on effectiveness and appropriateness of care, to provide vital feedback for improvements if necessary. In addition, the shorter travel time to HBOC can be explained because HBOC is more accessible, for example by public transportation, compared to the PC+ locations. Although PC+ focuses on care delivered closer to patients' homes, this does not guarantee a shorter travel time. This can be important for patients who rely on public transportation [48].

Despite the estimated PS balanced covariates for the overall study population, subgroups based on medical specialty showed large variability in covariates. Therefore, caution is advised in the interpretation of the HRQoL and experienced quality of care outcomes per medical specialty. Instead of a cohort study, a randomised controlled trial (RCT) with block randomisation could be a useful technique to achieve balance in the allocation of patients to subgroups and therewith reduce bias [49]. However, performing an RCT in this case was not possible and not preferable since the PC+ intervention was subject to change during the study period, with inflow and outflow of medical specialties, for example. Furthermore, an important principle in this intervention was that GPs remain responsible for the patient and therefore they decided in agreement with the patient whether to refer a patient to PC+.

There are several limitations to this study. Although PSM permits a more objective analysis by balancing the study groups with respect to confounders, it only allows for adjustment of measured confounders [37]. However, this limitation is applicable for all datasets and all multivariable adjustment methods. Sensitivity analysis was performed to assess the robustness of the study results. As the results changed minimally regarding statistical significance and direction of the association, confidence was provided that no significant unmeasured baseline characteristics were influencing the PC+ effect [50]. Only travel time turned out to be sensitive to the PSM method used. Furthermore, this study seems to be affected by non-responder bias since non-responders turned out to be significantly younger compared to responders [51]. Finally, this study was based on a single region with one primary care organisation and one hospital, which limits the generalisability of the results.

In conclusion, this study found equal results on HRQoL and experienced quality of care outcomes between patients referred to PC+ and HBOC. Therefore, it can be concluded that, despite the lack of diagnostic tools, population health and quality of care are maintained in PC+. In future research, there should be more emphasis on cost comparison for patients and for the total health system to demonstrate the potential added value of PC.

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ADDITIONAL FILES

Table S1 Baseline characteristics of responders and non-responders

	PC+		
Characteristics	**Responders**	**Non-responders**	**P-value**
N (%)	2,120 (54.5)	1,770 (45.5)	
Age (mean, SD) [A]	54.6 (16.32)	48.1 (17.06)	≤ 0.001 *
Gender (male) N (%) [B]	721 (40.8)	817 (38.5)	0.159
Medical specialty referred to [C]			0.716
Dermatology N (%)	661 (31.2)	565 (32.1)	
Gynaecology N (%)	126 (5.9)	92 (5.2)	
Internal medicine N (%)	58 (2.7)	55 (3.1)	
Otolaryngology N (%)	359 (16.9)	316 (18.0)	
Neurology N (%)	158 (7.5)	121 (6.9)	
Ophthalmology N (%)	178 (8.4)	144 (8.2)	
Orthopaedics N (%)	402 (19.0)	316 (18.0)	
Rheumatology N (%)	142 (6.7)	132 (7.5)	
Urology N (%)	26 (1.2)	15 (0.9)	

Note: PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SD = standard deviation

** P ≤ 0.001; A Age at date of appointment was missing for n=69 patients (PC+ group: n=2 non-responders and HBOC group: n=1 responders and n=66 non-responders); B Gender of n=1 patient was missing (PC+ group: n=1 responder); C Medical specialty referred to was missing for n=16 patients (PC+ group: n=10 non-responders and HBOC group: n=6 non-responders)*

	HBOC		
	Responders	**Non-responders**	**P-value**
	778 (47.3)	867 (52.7)	
	56.5 (15.79)	49.73 (17.34)	≤ 0.001 *
	337 (38.9)	314 (40.4)	0.537
			≤ 0.001 *
	127 (16.3)	119 (13.8)	
	55 (7.1)	102 (11.8)	
	71 (9.1)	91 (10.6)	
	103 (13.2)	88 (10.2)	
	96 (12.3)	148 (17.2)	
	62 (8.0)	42 (4.9)	
	188 (24.2)	205 (23.8)	
	30 (3.9)	28 (3.3)	
	46 (5.9)	38 (4.4)	

Table S2 Baseline characteristics before and after propensity score matching per medical specialty

Dermatology	Before PSM					
	PC+		HBOC		P-value	SMD
N	563		105			
Age (mean, SD)	54.85	54.85	56.96	16.72	0.238	0.126
Gender (male) (%, SD)	39%	39%	44%	0.50	0.382	0.092
Native country (Netherlands) (%, SD)	97%	97%	91%	0.28	0.003**	0.258
Educational level						
Low (%, SD)	18%	18%	20%	0.40	0.648	0.048
Medium (%, SD)	44%	44%	49%	0.50	0.432	0.083
High (%, SD)	37%	37%	31%	0.47	0.238	0.127
EQ-5D-5L (mean, SD)	0.88	0.88	0.86	0.17	0.29	0.104
EQ VAS (mean, SD)	81.02	81.02	77.05	17.18	0.008**	0.258
SF12 PCS (mean, SD)	51.24	51.24	48.87	10.43	0.007**	0.257
SF12 MCS (mean, SD)	52.05	52.05	51.4	9.20	0.485	0.073
BMI (mean, SD)	25.46	25.46	26.56	5.37	0.019*	0.228
Smoking behaviour						
Smoker (%, SD)	15%	15%	16%	0.37	0.849	0.020
Former smoker (%, SD)	42%	42%	44%	0.50	0.669	0.045
Non-smoker (%, SD)	43%	43%	40%	0.49	0.571	0.060
Alcohol user (%, SD)	63%	63%	64%	0.48	0.938	0.008
Gynaecology						
N	94		42			77
Age (mean, SD)	41.54	16.38	41.86	13.81	0.914	44.51
Gender (male) (%, SD)	0%	0.00	0%	0.00	-	0%
Native country (Netherlands) (%, SD)	91%	0.28	98%	0.15	0.187	92%
Educational level						
Low (%, SD)	4%	0.20	17%	0.38	0.014*	5%
Medium (%, SD)	56%	0.50	31%	0.47	0.006**	55%
High (%, SD)	39%	0.49	52%	0.51	0.159	40%
EQ-5D-5L(mean, SD)	0.89	0.13	0.85	0.20	0.137	0.89
EQ VAS (mean, SD)	82.88	13.76	80.17	13.55	0.287	82.21

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	210		105			
	58.4	15.55	56.96	16.72	0.453	0.089
	38%	0.49	44%	0.50	0.291	0.126
	97%	0.17	91%	0.28	0.025*	0.247
	21%	0.41	20%	0.40	0.770	0.035
	46%	0.50	49%	0.50	0.691	0.048
	32%	0.47	31%	0.47	0.865	0.020
	79.19	14.22	77.05	17.18	0.242	0.136
	0.86	0.14	0.86	0.17	0.901	0.014
	51.93	8.94	51.4	9.20	0.623	0.058
	49.62	8.43	48.87	10.43	0.497	0.078
	25.6	4.49	26.56	5.37	0.096	0.193
	20%	0.40	16%	0.37	0.473	0.087
	42%	0.49	44%	0.50	0.748	0.038
	39%	0.49	40%	0.49	0.807	0.029
	59%	0.49	64%	0.48	0.372	0.107
		42				
	15.77	41.54	16.38	41.86	13.81	
	0.00	0%	0.00	0%	0.00	
	0.27	91%	0.28	98%	0.15	
	0.22	4%	0.20	17%	0.38	
	0.50	56%	0.50	31%	0.47	
	0.49	39%	0.49	52%	0.51	
	0.14	0.89	0.13	0.85	0.20	
	13.89	82.88	13.76	80.17	13.55	

Table S2 Continued

Gynaecology	Before PSM					
	PC+		HBOC		P-value	SMD
SF12 PCS (mean, SD)	52.49	8.71	52.24	7.80	0.87	51.36
SF12 MCS (mean, SD)	51.03	9.45	47.39	9.00	0.037*	50.85
BMI (mean, SD)	25.42	4.50	24.73	3.67	0.386	25.91
Smoking behaviour						
Smoker (%, SD)	12%	0.32	21%	0.42	0.141	0.261
Former smoker (%, SD)	42%	42%	44%	0.50	0.669	0.045
Non-smoker (%, SD)	43%	43%	40%	0.49	0.571	0.060
Alcohol user (%, SD)	63%	63%	64%	0.48	0.938	0.008
Otolaryngology						
N	295		86			
Age (mean, SD)	58.73	14.25	59.07	14.14	0.845	0.024
Gender (male) (%, SD)	51%	0.50	43%	0.50	0.203	0.157
Native country (Netherlands) (%, SD)	97%	0.17	98%	0.15	0.725	0.045
Educational level						
Low (%, SD)	21%	0.41	19%	0.39	0.627	0.060
Medium (%, SD)	42%	0.49	35%	0.48	0.258	0.140
High (%, SD)	37%	0.48	47%	0.50	0.124	0.187
EQ-5D-5L (mean, SD)	0.84	0.14	0.84	0.12	0.809	0.031
EQ VAS (mean, SD)	76.22	16.12	72.77	16.32	0.082	0.213
SF12 PCS (mean, SD)	49.07	8.57	48.05	7.39	0.319	0.127
SF12 MCS (mean, SD)	51.02	8.97	49.86	9.57	0.297	0.126
BMI (mean, SD)	26.11	4.29	26.16	4.67	0.929	0.011
Smoking behaviour						
Smoker (%, SD)	14%	0.35	10%	0.31	0.367	0.114
Former smoker (%, SD)	45%	0.50	47%	0.50	0.773	0.035
Non-smoker (%, SD)	41%	0.49	43%	0.50	0.740	0.041
Alcohol user (%, SD)	69%	0.46	60%	0.49	0.148	0.175

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	9.13	52.49	8.71	52.24	7.80	
	9.63	51.03	9.45	47.39	9.00	
	4.72	25.42	4.50	24.73	3.67	
	13%	0.34	12%	0.32	21%	0.42
	42%	0.49	44%	0.50	0.748	0.038
	39%	0.49	40%	0.49	0.807	0.029
	59%	0.49	64%	0.48	0.372	0.107
	173		86			
	61.17	12.38	58.73	14.25	59.07	14.14
	50%	0.50	51%	0.50	43%	0.50
	97%	0.18	97%	0.17	98%	0.15
	24%	0.43	21%	0.41	19%	0.39
	35%	0.48	42%	0.49	35%	0.48
	41%	0.49	37%	0.48	47%	0.50
	0.84	0.13	0.84	0.14	0.84	0.12
	74.97	16.48	76.22	16.12	72.77	16.32
	48.05	8.81	49.07	8.57	48.05	7.39
	50.17	9.31	51.02	8.97	49.86	9.57
	26.31	4.26	26.11	4.29	26.16	4.67
	12%	0.33	14%	0.35	10%	0.31
	43%	0.50	45%	0.50	47%	0.50
	45%	0.50	41%	0.49	43%	0.50
	66%	0.47	69%	0.46	60%	0.49

Table S2 Continued

Otolaryngology	Before PSM					
	PC+		HBOC		P-value	SMD
N	295		86			
Age (mean, SD)	58.73	14.25	59.07	14.14	0.845	0.024
Gender (male) (%, SD)	51%	0.50	43%	0.50	0.203	0.157
Native country (Netherlands) (%, SD)	97%	0.17	98%	0.15	0.725	0.045
Educational level						
Low (%, SD)	21%	0.41	19%	0.39	0.627	0.060
Medium (%, SD)	42%	0.49	35%	0.48	0.258	0.140
High (%, SD)	37%	0.48	47%	0.50	0.124	0.187
EQ-5D-5L (mean, SD)	0.84	0.14	0.84	0.12	0.809	0.031
EQ VAS (mean, SD)	76.22	16.12	72.77	16.32	0.082	0.213
SF12 PCS (mean, SD)	49.07	8.57	48.05	7.39	0.319	0.127
SF12 MCS (mean, SD)	51.02	8.97	49.86	9.57	0.297	0.126
BMI (mean, SD)	26.11	4.29	26.16	4.67	0.929	0.011
Smoking behaviour						
Smoker (%, SD)	14%	0.35	10%	0.31	0.367	0.114
Former smoker (%, SD)	45%	0.50	47%	0.50	0.773	0.035
Non-smoker (%, SD)	41%	0.49	43%	0.50	0.740	0.041
Alcohol user (%, SD)	69%	0.46	60%	0.49	0.148	0.175
Internal medicine						
N	41		61			
Age (mean, SD)	52.54	18.46	54.74	16.19	0.526	0.127
Gender (male) (%, SD)	34%	0.48	36%	0.48	0.844	0.040
Native country (Netherlands) (%, SD)	98%	0.16	98%	0.13	0.778	0.056
Educational level						
Low (%, SD)	22%	0.42	13%	0.34	0.245	0.231
Medium (%, SD)	46%	0.50	57%	0.50	0.278	0.220
High (%, SD)	32%	0.47	30%	0.46	0.815	0.047
EQ-5D-5L (mean, SD)	0.74	0.22	0.78	0.14	0.244	0.227
EQ VAS (mean, SD)	67.63	17.37	70.03	15.90	0.473	0.144
SF12 PCS (mean, SD)	43.93	10.56	45.84	9.89	0.355	0.187
SF12 MCS (mean, SD)	46.28	10.28	48.57	10.33	0.274	0.222
BMI (mean, SD)	26.28	3.88	25.36	4.65	0.299	0.215

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	173		86			
	61.17	12.38	58.73	14.25	59.07	14.14
	50%	0.50	51%	0.50	43%	0.50
	97%	0.18	97%	0.17	98%	0.15
	24%	0.43	21%	0.41	19%	0.39
	35%	0.48	42%	0.49	35%	0.48
	41%	0.49	37%	0.48	47%	0.50
	0.84	0.13	0.84	0.14	0.84	0.12
	74.97	16.48	76.22	16.12	72.77	16.32
	48.05	8.81	49.07	8.57	48.05	7.39
	50.17	9.31	51.02	8.97	49.86	9.57
	26.31	4.26	26.11	4.29	26.16	4.67
	12%	0.33	14%	0.35	10%	0.31
	43%	0.50	45%	0.50	47%	0.50
	45%	0.50	41%	0.49	43%	0.50
	66%	0.47	69%	0.46	60%	0.49
	41		42			
	52.54	18.46	52.54	18.46	54.74	16.19
	34%	0.48	34%	0.48	36%	0.48
	98%	0.16	98%	0.16	98%	0.13
	22%	0.42	22%	0.42	13%	0.34
	46%	0.50	46%	0.50	57%	0.50
	32%	0.47	32%	0.47	30%	0.46
	0.74	0.22	0.74	0.22	0.78	0.14
	67.63	17.37	67.63	17.37	70.03	15.90
	43.93	10.56	43.93	10.56	45.84	9.89
	46.28	10.28	46.28	10.28	48.57	10.33
	26.28	3.88	26.28	3.88	25.36	4.65

Table S2 Continued

Internal medicine	Before PSM					
	PC+		HBOC		P-value	SMD
Smoking behaviour						
Smoker (%, SD)	12%	0.33	21%	0.41	0.241	0.244
Former smoker (%, SD)	37%	0.49	36%	0.48	0.958	0.011
Non- smoker (%, SD)	51%	0.51	43%	0.50	0.398	0.171
Alcohol user (%, SD)	63%	0.49	52%	0.50	0.278	0.221
Neurology						
N	131		78			
Age (mean, SD)	55.04	15.12	58.91	15.35	0.076	0.254
Gender (male) (%, SD)	42%	0.50	46%	0.50	0.559	0.084
Native country (Netherlands) (%, SD)	96%	0.19	96%	0.19	0.992	0.002
Educational level						
Low (%, SD)	17%	0.38	31%	0.46	0.018	0.331
Medium (%, SD)	53%	0.50	41%	0.50	0.104	0.234
High (%, SD)	31%	0.46	28%	0.45	0.723	0.051
EQ-5D-5L (mean, SD)	0.71	0.22	0.76	0.14	0.099	0.250
EQ VAS (mean, SD)	65.7	17.66	70.15	12.97	0.054	0.287
SF12 PCS(mean, SD)	43.18	8.93	42.67	9.04	0.693	0.057
SF12 MCS (mean, SD)	48.76	10.19	49.94	9.44	0.409	0.119
BMI (mean, SD)	26.4	4.76	26.52	4.45	0.857	0.026
Smoking behaviour						
Smoker (%, SD)	19%	0.39	21%	0.41	0.803	0.036
Former smoker (%, SD)	46%	0.50	40%	0.49	0.395	0.122
Non- smoker (%, SD)	35%	0.48	40%	0.49	0.505	0.095
Alcohol user (%, SD)	59%	0.49	62%	0.49	0.696	0.056
Ophthalmology						
N	156		55			
Age (mean, SD)	62.57	12.55	63.20	11.36	0.744	0.053
Gender (male) (%, SD)	44%	0.50	42%	0.50	0.821	0.036
Native country (Netherlands) (%, SD)	96%	0.19	96%	0.19	0.944	0.011

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P ‹ 0.05; ** P ‹ 0.01; *** P ‹ 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	12%	0.33	12%	0.33	21%	0.41
	37%	0.49	37%	0.49	36%	0.48
	51%	0.51	51%	0.51	43%	0.50
	63%	0.49	63%	0.49	52%	0.50
	126		75			
	55.77	14.84	55.04	15.12	58.91	15.35
	43%	0.50	42%	0.50	46%	0.50
	96%	0.20	96%	0.19	96%	0.19
	17%	0.38	17%	0.38	31%	0.46
	51%	0.50	53%	0.50	41%	0.50
	32%	0.47	31%	0.46	28%	0.45
	0.71	0.23	0.71	0.22	0.76	0.14
	65.59	17.83	65.7	17.66	70.15	12.97
	42.98	9.02	43.18	8.93	42.67	9.04
	48.43	10.23	48.76	10.19	49.94	9.44
	26.45	4.79	26.4	4.76	26.52	4.45
	20%	0.40	19%	0.39	21%	0.41
	46%	0.50	46%	0.50	40%	0.49
	34%	0.48	35%	0.48	40%	0.49
	58%	0.50	59%	0.49	62%	0.49
	120		55			
	63.72	11.87	62.57	12.55	63.20	11.36
	45%	0.50	44%	0.50	42%	0.50
	95%	0.22	96%	0.19	96%	0.19

Table S2 Continued

Ophthalmology	Before PSM					
	PC+		HBOC		P-value	SMD
Educational level						
Low (%, SD)	21%	0.41	24%	0.43	0.629	0.075
Medium (%, SD)	46%	0.50	40%	0.49	0.481	0.111
High (%, SD)	34%	0.48	36%	0.49	0.750	0.050
EQ-5D-5L (mean, SD)	0.87	0.14	0.85	0.16	0.534	0.095
EQ VAS (mean, SD)	77.81	14.00	80.07	13.45	0.299	0.165
SF12 PCS (mean, SD)	48.98	7.80	46.92	10.22	0.122	0.227
SF12 MCS (mean, SD)	52.24	7.99	52.81	9.63	0.668	0.064
BMI (mean, SD)	26.53	4.36	27.2	4.38	0.329	0.153
Smoking behaviour						
Smoker (%, SD)	17%	0.37	18%	0.39	0.798	0.040
Former smoker (%, SD)	54%	0.50	44%	0.50	0.195	0.204
Non- smoker (%, SD)	29%	0.46	38%	0.49	0.236	0.183
Alcohol user (%, SD)	64%	0.48	65%	0.48	0.858	0.028
Orthopaedics						
N	339		155			
Age (mean, SD)	57.35	13.47	60.66	13.02	0.011*	0.250
Gender (male) (%, SD)	40%	0.49	41%	0.49	0.961	0.005
Native country (Netherlands) (%, SD)	96%	0.21	95%	0.22	0.719	0.034
Educational level						
Low (%, SD)	19%	0.39	28%	0.45	0.021*	0.218
Medium (%, SD)	50%	0.50	54%	0.50	0.340	0.093
High (%, SD)	32%	0.47	18%	0.39	0.001**	0.322
EQ-5D-5L (mean, SD)	0.72	0.17	0.71	0.17	0.298	0.101
EQ VAS (mean, SD)	70.41	16.91	69.28	17.77	0.501	0.065
SF12 PCS (mean, SD)	41.64	8.77	40.29	9.77	0.126	0.146
SF12 MCS (mean, SD)	51.66	9.69	50.43	8.85	0.178	0.133
BMI (mean, SD)	27.06	4.64	27.34	4.84	0.533	0.060

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	22%	0.42	21%	0.41	24%	0.43
	45%	0.50	46%	0.50	40%	0.49
	32%	0.47	34%	0.48	36%	0.49
	0.86	0.13	0.87	0.14	0.85	0.16
	77.67	13.00	77.81	14.00	80.07	13.45
	48.04	8.18	48.98	7.80	46.92	10.22
	52.19	7.73	52.24	7.99	52.81	9.63
	26.87	4.25	26.53	4.36	27.2	4.38
	15%	0.36	17%	0.37	18%	0.39
	57%	0.50	54%	0.50	44%	0.50
	28%	0.45	29%	0.46	38%	0.49
	63%	0.48	64%	0.48	65%	0.48
	294		155			
	58.88	12.60	57.35	13.47	60.66	13.02
	40%	0.49	40%	0.49	41%	0.49
	95%	0.21	96%	0.21	95%	0.22
	21%	0.41	19%	0.39	28%	0.45
	47%	0.50	50%	0.50	54%	0.50
	33%	0.47	32%	0.47	18%	0.39
	0.72	0.16	0.72	0.17	0.71	0.17
	69.73	16.54	70.41	16.91	69.28	17.77
	40.91	8.56	41.64	8.77	40.29	9.77
	51.68	9.39	51.66	9.69	50.43	8.85
	27.32	4.74	27.06	4.64	27.34	4.84

Table S2 Continued

Orthopaedics	Before PSM					
	PC+		HBOC		P-value	SMD
Smoking behaviour						
Smoker (%, SD)	15%	0.36	15%	0.36	0.953	0.006
Former smoker (%, SD)	40%	0.49	46%	0.50	0.235	0.115
Non- smoker (%, SD)	45%	0.50	39%	0.49	0.254	0.111
Alcohol user (%, SD)	59%	0.49	55%	0.50	0.427	0.077
Rheumatology						
N	120		24			
Age (mean, SD)	55.08	13.32	58	14.15	0.334	0.212
Gender (male) (%, SD)	26%	0.44	50%	0.51	0.018*	0.507
Native country (Netherlands) (%, SD)	98%	0.13	100%	0.00	0.528	0.183
Educational level						
Low (%, SD)	26%	0.44	21%	0.41	0.609	0.117
Medium (%, SD)	50%	0.50	42%	0.50	0.459	0.166
High (%, SD)	24%	0.43	38%	0.49	0.178	0.288
EQ-5D-5L (mean, SD)	0.72	0.14	0.69	0.18	0.262	0.229
EQ VAS (mean, SD)	67.76	16.37	66.25	17.34	0.684	0.089
SF12 PCS (mean, SD)	41.50	8.60	40.68	8.64	0.668	0.096
SF12 MCS (mean, SD)	50.01	11.02	47.78	9.60	0.358	0.215
BMI (mean, SD)	26.55	4.68	27.04	7.11	0.671	0.081
Smoking behaviour						
Smoker (%, SD)	22%	0.41	29%	0.46	0.428	0.171
Former smoker (%, SD)	49%	0.50	33%	0.48	0.158	0.322
Non- moker (%, SD)	29%	0.46	38%	0.49	0.422	0.175
Alcohol user (%, SD)	57%	0.50	58%	0.50	0.881	0.033
Urology						
N	24		37			
Age (mean, SD)	56.25	19.09	55	15.61	0.781	0.072
Gender (male) (%, SD)	79%	0.41	62%	0.49	0.167	0.374
Native country (Netherlands) (%, SD)	100%	0.00	97%	0.16	0.425	0.232

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P ‹ 0.05; ** P ‹ 0.01; *** P ‹ 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	15%	0.36	15%	0.36	15%	0.36
	40%	0.49	40%	0.49	46%	0.50
	45%	0.50	45%	0.50	39%	0.49
	60%	0.49	59%	0.49	55%	0.50
	48		24			
	55.5	11.66	55.08	13.32	58	14.15
	19%	0.39	26%	0.44	50%	0.51
	96%	0.20	98%	0.13	100%	0.00
	31%	0.47	26%	0.44	21%	0.41
	52%	0.50	50%	0.50	42%	0.50
	17%	0.38	24%	0.43	38%	0.49
	0.69	0.15	0.72	0.14	0.69	0.18
	64.56	18.01	67.76	16.37	66.25	17.34
	39.04	9.37	41.50	8.60	40.68	8.64
	48.92	10.83	50.01	11.02	47.78	9.60
	26.71	4.38	26.55	4.68	27.04	7.11
	23%	0.42	22%	0.41	29%	0.46
	42%	0.50	49%	0.50	33%	0.48
	35%	0.48	29%	0.46	38%	0.49
	48%	0.50	57%	0.50	58%	0.50
	24		22			
	56.25	19.09	56.25	19.09	55	15.61
	79%	0.41	79%	0.41	62%	0.49
	100%	0.00	100%	0.00	97%	0.16

Table S2 Continued

Urology	Before PSM					
	PC+		HBOC		P-value	SMD
Educational level						
Low (%, SD)	29%	0.46	16%	0.37	0.235	0.307
Medium (%, SD)	50%	0.51	43%	0.50	0.612	0.133
High (%, SD)	21%	0.41	41%	0.50	0.113	0.430
EQ-5D-5L (mean, SD)	0.82	0.20	0.85	0.14	0.568	0.145
EQ VAS (mean, SD)	72.79	19.45	75.68	11.92	0.475	0.179
SF12 PCS (mean, SD)	49.14	6.96	48.72	8.40	0.840	0.054
SF12 MCS (mean, SD)	49.90	10.29	49.2	8.25	0.768	0.076
BMI (mean, SD)	27.47	5.26	25.04	3.97	0.045*	0.520
Smoking behaviour						
Smoker (%, SD)	12%	0.34	14%	0.35	0.911	0.030
Former smoker (%, SD)	46%	0.51	46%	0.51	0.993	0.002
Non- smoker (%, SD)	42%	0.50	41%	0.50	0.932	0.022
Alcohol user (%, SD)	50%	0.51	62%	0.49	0.356	0.243

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P < 0.05; ** P < 0.01; *** P < 0.001*

	After PSM					
	PC+		HBOC		P-value	SMD
	29%	0.46	29%	0.46	16%	0.37
	50%	0.51	50%	0.51	43%	0.50
	21%	0.41	21%	0.41	41%	0.50
	0.82	0.20	0.82	0.20	0.85	0.14
	72.79	19.45	72.79	19.45	75.68	11.92
	49.14	6.96	49.14	6.96	48.72	8.40
	49.9	10.29	49.90	10.29	49.2	8.25
	27.47	5.26	27.47	5.26	25.04	3.97
	12%	0.34	12%	0.34	14%	0.35
	46%	0.51	46%	0.51	46%	0.51
	42%	0.50	42%	0.50	41%	0.50
	50%	0.51	50%	0.51	62%	0.49

Table S3 Health-related quality of life outcomes before and after propensity score matching per medical specialty

	Dermatology		**Gynaecology**	
	BeforePSM	After PSM	Before PSM	After PSM
EQ-5D-5L				
Time T1 x study group [A]	-	-	-	-
Time T2 x study group [A]	-	-	-	-
EQ-VAS				
Time T1 x study group [A]	-	-	-	-
Time T2 x study group [A]	-	-	-	-
SF-12 PCS				
Time T2 x study group [A]	HBOC *	HBOC *	-	-
SF-12 MCS				
Time T2 x study group [A]	-	-	-	-
PGIC B				
Time T2 x study group [A]	-	-	-	-
	Ophthalmology		**Orthopaedics**	
	Before PSM	After PSM	Before PSM	After PSM
EQ-5D-5L				
Time T1 x study group [A]	-	-	-	-
Time T2 x study group [A]	PC+ *	-	-	-
EQ-VAS				
Time T1 x study group [A]	-	-	-	-
Time T2 x study group [A]	-	-	-	-
SF-12 PCS				
Time T2 x study group [A]	-	-	-	-
SF-12 MCS				
Time T2 x study group [A]	-	-	-	-
PGIC B				-
Time T2 x study group [A]	-	-	-	-

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; In table: PC+ = significant higher scores in the PC+ group; HBOC = significant higher scores in the HBOC group

** P < 0.05; [A] Group was coded as 1 = HBOC group and 0 = PC+ group; [B] PGIC was measured at T1 and T2, not at baseline*

	Otolaryngology		**Internal medicine**		**Neurology**	
	Before PSM	After PSM	Before PSM	After PSM	BeforePSM	AfterPSM
	-	-	-	-	PC+*	PC+ *
	-	-	-	-	PC+*	-
	-	-	-	-	PC+*	PC+ *
	-	-	-	-	PC+*	PC+ *
	-	-	-	-	-	-
	-	PC+ *	-	PC+ *	-	-
	PC+ *	PC+ *	-	-	-	-
	Rheumatology		**Urology**			
	Before PSM	After PSM	Before PSM	After PSM		
	-	-	-	-		
	-	-	-	-		
	-	-	-	-		
	-	-	-	-		
	-	-	-	-		
	-	-	-	-		
	-	-	-	-		
	-	-	-	-		

Table S4 Comparison of patient experienced quality of care outcomes before and after propensity score matching per medical specialty

	Dermatology	
	Before PSM	**After PSM**
Timeliness (1)		
Waiting time for appointment	-	-
Waiting time in waiting room ‹30 minutes	PC+ **	PC+ **
Treatment by the medical specialist		
Complaint was taken seriously	-	-
Specialist listened carefully	-	-
Specialist took enough time	-	-
Treated with respect	-	-
Competence of the specialist	-	-
Overall help of the specialist	-	-
Information provision and communication by the medical specialist		
Information about different treatment options	-	-
Understandable explanation	-	-
Opportunity to ask questions	-	PC+ *
Shared decision making	-	-
Communication and collaboration between the GP and medical specialist		
Matching recommendations between GP and specialist	-	-
Awareness of the medical specialist about the complaint	-	-
Collaboration and alignment between GP and specialist	-	-
Overall assessment of quality of care (1)		
Recommend medical specialist to family/friends	-	-
Recommend PC+/HBOC to family/friends	-	-
Timeliness(2)		
Travel time (in minutes) [A]	HBOC **	HBOC **
Overall assessment of quality of care (2)		
Grade specialist (0-10)	-	-
Grade PC+/HBOC (0-10)	PC+ **	PC+ *

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; In table: PC+ = significant higher scores in the PC+ group; HBOC = significant higher scores in the HBOC group

** P ‹ 0.01; ** P ‹ 0.00; [A] A significant higher score on travel time means a longer travel time in minutes and is in this case a unfavourable result*

	Gynaecology		Otolaryngology		Internal medicine		Neurology	
	Before PSM	After PSM	BeforePSM	After PSM	Before PSM	After PSM	Before PSM	After PSM
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	PC+ *	PC+ *
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	HBOC **	HBOC*	-	-	-	-
	-	-	-	-	-	-	-	-
	-	-	-	-	-	-	-	-

Table S4 Continued

	Ophthalmology	
	Before PSM	**AfterPSM**
Timeliness (1)		
Waiting time for appointment	-	-
Waiting time in waiting room <30 minutes	-	-
Treatment by the medical specialist		
Complaint was taken seriously	-	-
Specialist listened carefully	-	-
Specialist took enough time	-	-
Treated with respect	-	-
Competence of the specialist	-	-
Overall help of the specialist	-	-
Information provision and communication by the medical specialist		
Information about different treatment options	-	-
Understandable explanation	PC+*	-
Opportunity to ask questions	-	-
Shared decision making	-	-
Communication and collaboration between the GP and medical specialist		
Matching recommendations between GP and specialist	-	-
Awareness of the medical specialist about the complaint	-	-
Collaboration and alignment between GP and specialist	-	-
Overall assessment of quality of care (1)		
Recommend medical specialist to family/friends	-	-
Recommend PC+/HBOC to family/friends	-	-
Timeliness (2)		
Travel time (in minutes) [A]	-	-
Overall assessment of quality of care (2)		
Grade specialist (0-10)	-	-
Grade PC+/HBOC (0-10)	-	-

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; In table: PC+ = significant higher scores in the PC+ group; HBOC = significant higher scores in the HBOC group

** P < 0.01; ** P < 0.00; [A] A significant higher score on travel time means a longer travel time in minutes and is in this case a unfavourable result*

Orthopaedics		Rheumatology		Urology	
Before PSM	AfterPSM	Before PSM	After PSM	Before PSM	After PSM
-	-	-	-	PC+*	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-
HBOC**	HBOC**	-	-	-	-
-	-	-	-	-	-
-	-	-	-	-	-

Table S5 Baseline characteristics before and after propensity score matching

	Before PSM					
	PC+		HBOC		P-value	SMD
N	**1763**		**643**			
Age (mean, SD)	55.95	15.68	57.63	15.23	0.019*	0.109
Gender (male) (%, SD)	39%	0.49	41%	0.49	0.557	0.027
Native country (Netherlands) (%, SD)	97%	0.18	96%	0.20	0.393	0.038
Educational level						
Low (%, SD)	19%	0.39	22%	0.42	0.063	0.084
Medium (%, SD)	47%	0.50	46%	0.50	0.593	0.025
High (%, SD)	34%	0.48	32%	0.47	0.317	0.046
EQ-5D-5L (mean, SD)	0.81	0.17	0.79	0.17	0.005**	0.129
EQ VAS (mean, SD)	75.53	16.32	73.08	16.31	0.001**	0.150
SF12 PCS (mean, SD)	47.44	9.33	45.39	10.04	≤0.001***	0.211
SF12 MCS (mean, SD)	51.22	9.35	50.11	9.34	0.010**	0.119
BMI (mean, SD)	26.20	4.60	26.50	4.92	0.170	0.062
Smoking behaviour						
Smoker (%, SD)	16%	0.36	17%	0.38	0.443	0.035
Former smoker (%, SD)	42%	0.49	42%	0.49	0.771	0.013
Non-smoker (%, SD)	42%	0.49	41%	0.49	0.781	0.013
Alcohol user (%, SD)	62%	0.48	60%	0.49	0.198	0.059
Medical specialty referred to						
Dermatology (%, SD)	32%	0.47	16%	0.37	≤0.001***	0.371
Gynaecology (%, SD)	5%	0.22	7%	0.25	0.260	0.051
Internal medicine (%, SD)	2%	0.15	9%	0.29	‹0.001***	0.307
Otolaryngology (%, SD)	17%	0.37	13%	0.34	0.046	0.094
Neurology (%, SD)	7%	0.26	12%	0.33	≤0.001***	0.159
Ophthalmology (%, SD)	9%	0.28	9%	0.28	0.821	0.010
Orthopaedics (%, SD)	19%	0.39	24%	0.43	0.009**	0.119
Rheumatology (%, SD)	7%	0.25	4%	0.19	0.005**	0.138
Urology (%, SD)	1%	0.12	6%	0.23	≤0.001***	0.239

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation

** P ‹ 0.05; ** P ‹ 0.01; *** P ‹ 0.000*

	After PSM					
	PC+		HBOC		P-value	SMD
	608		608			
	57.25	15.10	57.81	15.18	0.516	0.037
	43%	0.50	41%	0.49	0.450	0.043
	95%	0.21	96%	0.20	0.679	0.024
	21%	0.41	22%	0.42	0.677	0.024
	46%	0.50	46%	0.50	0.954	0.003
	33%	0.47	32%	0.47	0.759	0.018
	0.79	0.17	0.79	0.17	0.866	0.010
	73.17	16.76	73.03	16.38	0.883	0.008
	45.73	9.78	45.34	10.06	0.489	0.040
	49.97	9.59	50.23	9.39	0.628	0.028
	26.54	4.65	26.53	4.82	0.984	0.001
	16%	0.37	17%	0.38	0.758	0.018
	45%	0.50	42%	0.49	0.326	0.056
	39%	0.49	41%	0.49	0.447	0.044
	61%	0.49	59%	0.49	0.482	0.040
	17%	0.38	17%	0.38	0.940	0.004
	8%	0.27	7%	0.25	0.511	0.038
	7%	0.25	7%	0.26	0.733	0.020
	13%	0.34	14%	0.35	0.740	0.019
	13%	0.33	12%	0.33	0.931	0.005
	9%	0.28	9%	0.29	0.762	0.017
	26%	0.44	25%	0.44	0.896	0.008
	4%	0.19	4%	0.19	1.000	0.001
	4%	0.19	4%	0.19	1.000	0.001

Table S6 Health-related quality of life outcomes before and after propensity score matching

	Before PSM		
EQ-5D-5L	**Estimate**	**SE**	**95% CI**
Intercept	0.82***	0.00	0.81 - 0.82
Study group [A]	-0.02**	0.01	-0.04 - -0.01
Time T1	0.01***	0.00	0.01 - 0.02
Time T2	0.02***	0.00	0.01 - 0.02
Time T1 x study group	-0.01*	0.01	-0.02 - 0.00
Time T2 x study group	-0.01*	0.01	-0.03 - -0.00
EQ-VAS	**Estimate**	**SE**	**95% CI**
Intercept	75.53***	0.39	74.77 – 76.29
Study group [A]	-2.45**	0.75	-3.92 – -0.97
Time T1	0.92**	0.31	0.32 – 1.53
Time T2	1.10	0.64	-0.16 – 2.35
Time T1 x study group	-0.41	0.60	-1.58 – 0.77
Time T2 x study group	-0.16	1.25	-2.60 – 2.28
SF-12 PCS	**Estimate**	**SE**	**95% CI**
Intercept	47.44***	0.23	46.99 – 47.88
Study group [A]	-2.05***	0.44	-2.91 – -1.19
Time T2	0.57***	0.17	0.24 – 0.90
Time T2 x study group	0.11	0.33	-0.53 – 0.75
SF-12 MCS	**Estimate**	**SE**	**95% CI**
Intercept	51.22***	0.22	50.78 – 51.66
Study group [A]	-1.11**	0.43	-1.95 – -0.27
Time T2	0.03	0.20	-0.36 – 0.42
Time T2 x study group	-0.32	0.39	-1.08 – 0.44
PGIC [B]	**Estimate**	**SE**	**95% CI**
Intercept	4.57***	0.73	3.14 – 6.01
Study group [A]	-1.10	1.41	-3.88 – 1.67
Time T2	-1.58*	0.73	-3.02 - -0.15
Time T2 x study group	1.11	1.42	-1.67 – 3.89

Note: PSM = propensity score matching; SE = standard error; CI = confidence interval

** P < 0.05; ** P < 0.01; *** P < 0.00; [A] Group was coded as 1 = Hospital Based Outpatient Care (HBOC) group and 0 = Primary Care Plus (PC+) group; [B] PGIC was measured at T1 and T2, not at baseline*

After PSM		
Estimate	**SE**	**95% CI**
0.79***	0.01	0.78 – 0.81
-0.00	0.01	-0.02- 0.02
0.01	0.01	0.00 – 0.03
0.01*	0.00	0.00 – 0.02
-0.01	0.01	-0.03 – 0.01
-0.01	0.01	-0.02 – 0.00
Estimate	**SE**	**95% CI**
73.17***	0.67	71.85 – 74.49
-1.14	0.95	-2.00 – 1.72
0.84	0.48	-0.11 – 1.79
1.29*	0.60	0.10 – 2.47
-0.39	0.68	-1.73 – 0.95
-0.61	0.86	-2.29 – 1.06
Estimate	**SE**	**95% CI**
45.73***	0.40	44.94 – 46.52
-0.40	0.57	-1.51 – 0.72
1.31***	0.29	0.74 – 1.87
-0.63	0.41	-1.44 – 0.17
Estimate	**SE**	**95% CI**
49.97***	0.38	49.21 – 51.72
0.26	0.54	-0.80 – 1.33
0.22	0.35	-0.37 – 1.00
-0.74	0.50	-1.71 – 0.23
Estimate	**SE**	**95% CI**
3.44***	0.04	3.36 – 3.51
0.05	0.05	-0.05 – 0.16
-0.35***	0.05	-0.46 - -0.24
-0.15	0.08	-0.30 – 0.00

Table S7 Comparison of patient experienced quality of care outcomes before and after propensity score matching

	Before PSM
	PC+
N	**1741**
Quality of care domains	**Satisfied/Yes n (%)**
Timeliness (1)	
Waiting time for appointment	89.2(1527)
Waiting time in waiting room <30 minutes	93.5 (1605)
Treatment by the medical specialist	
Complaint was taken seriously	97.5(1672)
Specialist listened carefully	97.3(1667)
Specialist took enough time	98.0(1679)
Treated with respect	98.8(1692)
Competence of the specialist	98.4(1673)
Overall help of the specialist	94.2(1612)
Information provision and communication by the medical specialist	
Information about different treatment options	92.6(1581)
Understandable explanation	97.1(1663)
Opportunity to ask questions	97.4(1666)
Shared decision making	88.4(892)
Communication and collaboration between the GP and medical specialist	
Matching recommendations between GP and specialist	80.3(1357)
Awareness of the medical specialist about the complaint	89.4(1519)
Collaboration and alignment between GP and specialist	85.8(1366)
Overall assessment of quality of care (1)	
Recommend medical specialist to family/friends	93.7(1598)
Recommend PC+/HBOC to family/friends	95.4(1625)
	Mean (SD)
Timeliness (2)	
Travel time (in minutes) [A]	15.6 (9.34)
Overall assessment of quality of care (2)	
Grade specialist (0-10)	8.5 (1.15)
Grade PC+/HBOC (0-10)	8.5 (1.08)

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; GP = general practitioner; SD = standard deviation

** P < 0.01; ** P < 0.00; [A] A significant higher score on travel time means a longer travel time in minutes and is in this case a unfavourable result*

	HBOC	
	624	
	Satisfied/Yes n (%)	P-value
	86.0(533)	0.032
	88.5 (546)	≤0.001**
	97.4(601)	0.845
	97.4(601)	0.845
	98.7(608)	0.240
	98.5(607)	0.574
	98.0(601)	0.543
	93.5(575)	0.553
	90.7(555)	0.140
	96.1(592)	0.236
	96.4(594)	0.231
	87.3(338)	0.582
	82.1(501)	0.324
	89.1(547)	0.827
	81.6(482)	0.016
	92.5(568)	0.298
	93.8(577)	0.119
	Mean (SD)	
	19.2 (12.18)	≤0.001**
	8.4 (1.22)	0.007*
	8.3 (1.11)	≤0.001**

Table S7 Continued

	After PSM
	PC+
N	**600**
Quality of care domains	**Satisfied/Yes n (%)**
Timeliness (1)	
Waiting time for appointment	90.2(535)
Waiting time in waiting room <30 minutes	91.9(545)
Treatment by the medical specialist	
Complaint was taken seriously	98.1(583)
Specialist listened carefully	98.3(584)
Specialist took enough time	98.5(585)
Treated with respect	99.2(589)
Competence of the specialist	99.2(586)
Overall help of the specialist	94.8(562)
Information provision and communication by the medical specialist	
Information about different treatment options	92.6(550)
Understandable explanation	97.0(576)
Opportunity to ask questions	97.6(579)
Shared decision making	89.5(315)
Communication and collaboration between the GP and medical specialist	
Matching recommendations between GP and specialist	80.6(473)
Awareness of the medical specialist about the complaint	89.3(527)
Collaboration and alignment between GP and specialist	85.6(476)
Overall assessment of quality of care (1)	
Recommend medical specialist to family/friends	94.9(563)
Recommend PC+/HBOC to family/friends	95.8(568)
	Mean (SD)
Timeliness (2)	
Travel time (in minutes) [A]	16.3 (9.29)
Overall assessment of quality of care (2)	
Grade specialist (0-10)	8.5 (1.20)
Grade PC+/HBOC (0-10)	8.5 (1.06)

Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; GP = general practitioner; SD = standard deviation

** P < 0.01; ** P < 0.00; [A] A significant higher score on travel time means a longer travel time in minutes and is in this case a unfavourable result*

	HBOC	
	589	
	Satisfied/Yes n (%)	**P-value**
	86.5(507)	0.047
	88(514)	0.026
	97.3(568)	0.309
	97.3(568)	0.217
	98.6(575)	0.837
	98.5(574)	0.267
	98.1(569)	0.122
	93.3(543)	0.286
	90.3(523)	0.165
	95.9(559)	0.315
	96.2(561)	0.160
	86.7(320)	0.252
	82.0(473)	0.165
	89.5(520)	0.921
	81.5(455)	0.067
	92.1(535)	0.047
	93.8(546)	0.128
	Mean (SD)	
	16.8 (11.18)	0.212
	8.4 (1.14)	0.048
	8.4 (1.11)	0.014

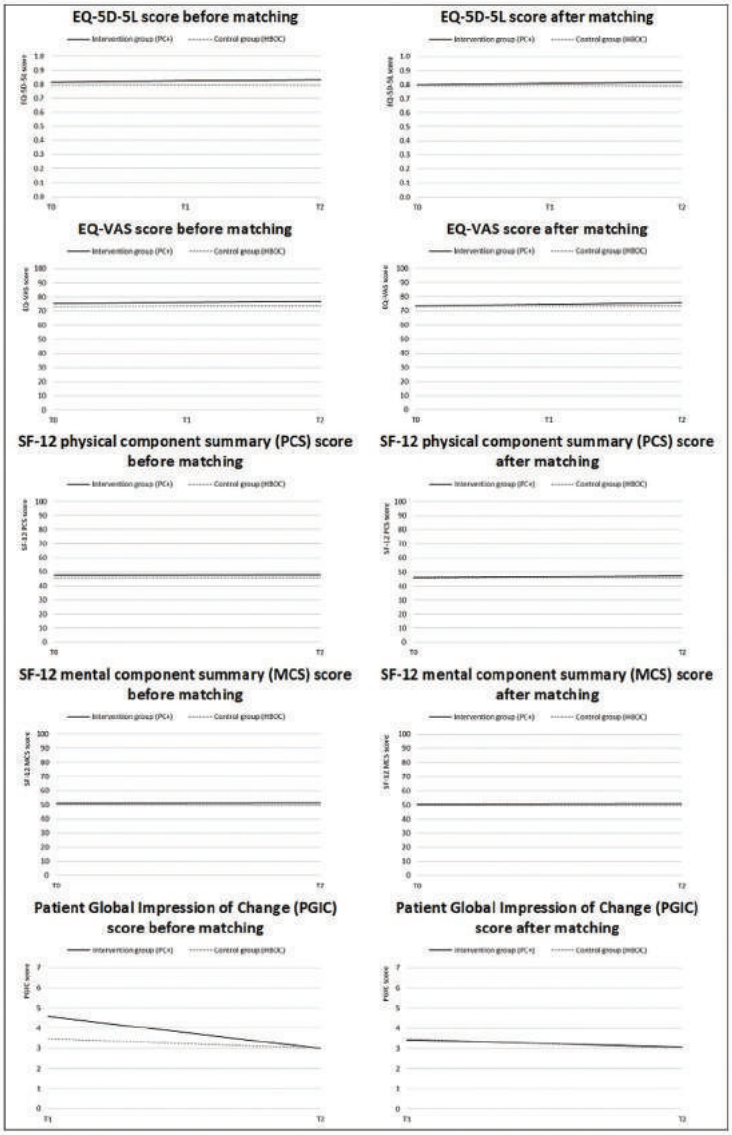
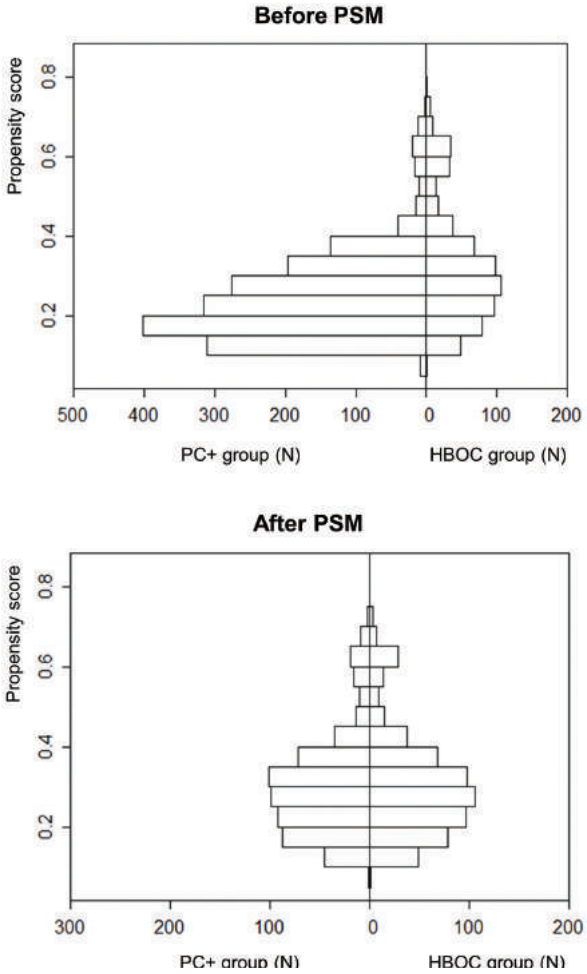


Figure S1 Uncorrected and Corrected Health-related Quality of Life Outcomes



Note: PSM = propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

Figure S2 Overlap of the Propensity Score in the Two Study Groups

06

Economic evaluation of new models of care

Does the decision change between Cost-Utility Analysis and Multi-Criteria Decision Analysis?

This chapter is submitted as:

van den Bogaart EHA, Kroese, MEAL, Spreeuwenberg MD, Ruwaard D, Tsiachristas A. Economic evaluation of new models of care: does the decision change between Cost-Utility Analysis and Multi-Criteria Decision Analysis?

ABSTRACT

Objectives: To experiment with new approaches of collaboration in healthcare delivery, local authorities implement new models of care. Regarding the local decision-context of these models, Multi-Criteria Decision Analysis (MCDA) may be of added value to Cost-Utility Analysis (CUA), as it covers a wider range of outcomes. This study compares the two methods using a side-by-side application.

Methods: A new Dutch model of care, Primary Care Plus (PC+), was used as case study to compare the results of CUA and MCDA. Data of patients referred to PC+ or care-as-usual were retrieved by questionnaires and administrative databases with a three-month follow-up. Propensity score matching together with generalised linear regression models was used to reduce confounding. Univariate and probabilistic sensitivity analyses were performed to explore uncertainty in the results.

Results: Although both methods indicated PC+ as dominant alternative, complementary differences were observed. MCDA provided additional evidence that PC+ improved access to care (standardised performance score of 0.742 versus 0.670), improvement in health-related quality of life was driven by the psychological well-being component (standardised performance score of 0.710 versus 0.704), estimated the budget required for the PC+ to be affordable in addition to preferable (€521.42 per patient). Additionally, MCDA was less sensitive to the utility measures used.

Conclusions: MCDA may facilitate an auditable and transparent evaluation of new models of care by providing additional information on a wider range of outcomes and incorporating affordability. However, more effort is needed to increase the usability of MCDA among local decision makers.

INTRODUCTION

New models of care are implemented worldwide to increase accessibility, equitability and affordability of care by developing new approaches of collaboration and delivering health and social care [1]. Health authorities are experimenting with new models of care at local level (so-called pilot sites, pioneer sites, or vanguards) that act as blueprints and inspiration to the rest of the healthcare system [2]. These models may take different forms as they are tailored to the local needs and context and are driven by local leaders from multiple-organisations who collaborate and are responsible to improve access and quality of care for their local populations [3-6]. However, learning from and scaling-up these local initiatives at a national level is challenging given the lack of a framework that assesses their cost-effectiveness [7, 8].

The appropriateness of the widely accepted Cost-Utility Analysis (CUA) in local decision-making is debatable. This is because the decision context is different than in health interventions and technologies that are subject to reimbursement decisions or national clinical guidelines [9, 10]. Furthermore, CUA includes quality-adjusted life-year (QALY) as a single measure of outcome and fails to incorporate outcomes of interest to local decision makers such as access to care, equity, patient satisfaction, and non-health related quality of life [11-13]. It is widely argued that economic evaluations of new models of care should be carried out at a local level [13, 14], should be flexible to accommodate the selection of all relevant outcomes and costs at different levels (e.g. individual, organisational, local) [15-18], and should incorporate the perspective of all relevant stakeholders [19]. Local decision makers are often using decision support tools, such as balanced scorecards and key performance indicators, to monitor various performances. However, these tools, similar to a Cost-Consequence Analysis (CCA), lack of clear decision rules to indicate cost-effectiveness [20, 21].

Multi-Criteria Decision Analysis (MCDA) is proposed as a suitable method for evaluating new models of care based on the performance on all relevant outcomes and perspectives of all relevant stakeholders [22-24]. In previous studies, MCDA was used to evaluate integrated care initiatives [25, 26]. However, it is unclear if the adoption of MCDA in the evaluation of new models of care would alter funding decisions at local level. Therefore, this paper aims to investigate whether the adoption of MCDA instead of traditional CUA alters the decision of investing in new models of care by using a Dutch new model of care as a case study. By conducting a side-by-side application of the CUA and MCDA, the applicability and suitability of

both methods to support local decision makers about the broader value for money of new model of care is tested.

METHODS

Setting and decision context

Primary Care Plus (PC+), was a new model of care implemented in the Dutch pioneer site Blue Care in the Southern Netherlands in 2014 by primary and secondary care providers, local health authorities, the largest health insurer in the region, and a local patient organisation [27, 28]. Its triple aim was to improve population health and patient satisfaction by improving quality of and access to care while avoiding unnecessary outpatient hospital visits [29]. After its piloting, the decision of the multiple collaborating and responsible organisations (i.e. stakeholders) was to disinvest or scale-up its implementation in other areas in the Netherlands.

Primary Care Plus versus Hospital Based Outpatient Care

PC+ was compared to care-as-usual, which was hospital-based outpatient care (HBOC). GPs within the pioneer site Blue Care were able to refer non-acute and low-complex patients either to a medical specialist in one of the two PC+ centres in the city of Maastricht or to HBOC. Despite the fact that PC+ was available for all GPs within the region, only one-third of the GPs referred patients regularly.

In PC+, the medical specialist examined and/or treated the patient during a maximum of two consultations. Following PC+, the medical specialist referred the patient back to the GP with treatment advice, or, if necessary, referred the patient to HBOC for further diagnosis and/or treatment. PC+ consultations were provided by medical specialists that worked on the HBOC in the Maastricht UMC+ who visited PC+ centres on a regular basis (weekly or biweekly).

Study design and data

In this longitudinal prospective observational study with a three-month follow-up, we used data of 2,116 adult patients who visited initially PC+ (intervention group) or HBOC (care-as-usual group) between December 2014 and April 2018. Data on patients' health and wellbeing and experience of care were collected prospectively using a survey. Healthcare consumption and related costs in PC+ and hospital care were retrieved retrospectively from patient medical records. The different data sources were linked and merged into one dataset. Diagnosis Treatment Combinations

(DTCs) at baseline were used to select patients referred to HBOC for low-complex hospital care (one or two consultations with a medical specialist) to define a control group comparable to the PC+ group.

Outcomes

Outcomes (or criteria in the MCDA context) were selected and grouped according to the Triple Aim [30] (i.e. health and well-being, experience of care, and costs) following Stiefel and Nolan's overview of outcomes per aim [31] and were operationalised using indicators (i.e. outcome measurements). A brief description of the outcomes related to the Triple Aim and the associated indicators is provided below and presented in Table 1. A more detailed description of the indicators is included in Table S1.

Self-reported health and well-being was measured in patients using the EuroQol five dimensional questionnaire with five levels (EQ-5D-5L) and the Short Form Health Survey version 2 (SF-12v2) at baseline, one-week and three-month follow-up. The EQ-5D-5L is a generic measure based on the construct of Health-Related Quality of Life and consists of questions on mobility, self-care, pain/discomfort, usual activities and anxiety/depression [32]. The SF-12v2 is focusing on the outcomes of 'physical functioning' and 'psychological well-being' [33]. Both indicators can be applied to persons with all different types of diseases and complaints. The two criteria (i.e. person-centeredness and access to care) related to the experience of care were measured using the Consumer Quality (CQ) index at one-week follow-up. The CQ index is a scientifically based, standardised tool, which can be used throughout the care sector to measure self-reported experience of care from the patient perspective [34]. For person-centeredness, we used a multi-component indicator based on three items of the CQ index, namely the degree that care matches an individual's needs, capabilities and preferences, and jointly making informed decisions. Furthermore, we used the time between referral and start of treatment as the indicator of the criterion access to care. The indicator for the criterion cost of care were the costs of all consultations with a medical specialist (including at PC+) and all hospital admissions form baseline to three-month follow-up.

Table 1 Criteria and related indicators relevant for Primary Care Plus

Triple Aim	Criteria	Indicators
Health and wellbeing	Health related quality of life	EuroQol five-dimensional questionnaire with five levels (EQ-5D-5L)
	Physical functioning	Short-Form Health Survey version 2 (SF-12v2)
	Psychological well-being	Short-Form Health Survey version 2 (SF-12v2)
Experience of care	Person-centeredness	Consumer Quality (CQ) index
	Access to care	Consumer Quality (CQ) index
Costs of care	Costs of care	Healthcare consumption and related costs in Primary Care Plus and hospital care

Note: all data were collected through questionnaires except for costs that were retrieved from administrative databases

Statistical analysis and propensity score matching

Descriptive statistics were produced in terms of means and standard deviations (SDs) for continuous variables, and frequencies for categorical variables. Moreover, propensity score matching (PSM) was performed to reduce observed confounding between the two groups by following a stepwise strategy [35-37]. First, we followed standard practice [38] and included in the propensity score model all possible confounding variables available in the dataset: age, gender, residence at birth, educational level, baseline health status (SF-12v2 physical and mental component summary score), historical healthcare costs (12 months before baseline) and medical specialty referred to. Second, the two groups were matched using several PSM techniques, including exact matching, Nearest Neighbor greedy, caliper (0.25) and optimal matching, full matching, genetic matching and inverse probability weighting. In addition to 1-to-1 matching, n-to-1 matching was used to keep a larger sample size since the HBOC group was smaller. The performance of the different PSM techniques on covariate balancing was assessed based on standardised mean differences (SMDs), Rubin's B (the absolute standardised difference of the means of the linear index of the propensity score in the intervention and (matched) control group) and Rubin's R (the ratio of intervention to (matched) control variances of the propensity score index) (see Table S2) [39]. SMDs of < 0.25, Rubin's B < 25 and Rubin's R between 0.5 and 2 indicate sufficient balance between the two groups. The PSM technique with the lowest values on these performance indicators was chosen and compared with the covariate balance before PSM.

Furthermore, we produced a doubly robust estimation to further reduce confounding by fitting generalised linear regression models to the complete cases and including the potential confounders and weights from the PSM in the regression [40]. In the regressions with QALYs as dependent variable, we also included patients' baseline utility as suggested in the literature [41]. The distribution family and link function in each regression were selected based on goodness-of-fit using the Akaike's information criterion (AIC) and the Bayesian Information Criterion (BIC) [42-44]. All statistical analyses were performed in R, an open source statistical programming environment (R Core Team 2016).

Cost-Utility Analysis

In the CUA, the cost of care related to PC+ and hospital care were used. QALYs were calculated using the area under the curve approach based on the EuroQol five-dimensional questionnaire (EQ-5D-5L) and Dutch value set [46, 47]. After performing

the doubly robust estimation by using generalised linear regression models, the differences between the intervention and control group were expressed by calculating the incremental cost-effectiveness ratios (ICERs) [48].

Multi-Criteria Decision Analysis

The MCDA included all criteria (i.e. outcomes) presented in Table 1 except of the EQ-5D-5L, since patients' health and wellbeing was already covered by the SF-12v2. Including the EQ-5D-5L next to the SF-12v2 would cause overlap of criteria and therefore double counting [49]. Cost calculation and doubly robust estimation was performed similar to the CUA. As the criteria were measured on different scales, we standardised them using the relative standardisation method (details on this method are presented in Appendix 1). Criteria weights (i.e. the relative importance of each outcome) were retrieved from the SELFIE study, which estimated weights of Dutch stakeholders for new models of care [26, 50]. The weights for all criteria were derived from the exact same criteria used in the SELFIE study except the criterion access of care for which we used the weight of the "continuity of care" criterion in the SELFIE study. Finally, the standardised performance scores were combined with the weights and converted into an overall value for the PC+ intervention and HBOC according to the value-based Multi-Attribute Utility Theory (MAUT) method, using a 'weighted sum approach' [49, 51, 52].

Sensitivity analyses

In both the CUA and MCDA, probabilistic sensitivity analysis using Monte-Carlo simulation, with 5,000 replications, was performed to address second order uncertainty in the results [53, 54]. Regarding the CUA, the confidence interval around the ICER was calculated using bootstrapped estimations of the mean cost and QALY differences [55]. This was graphically presented using a cost-effectiveness acceptability curve (CEAC) [56]. The uncertainty in the MCDA was graphically presented using a Conditional Multi-attribute Acceptability Curve (CMAC) [26]. In contrast to the CEAC, the probability of PC+ being cost-effective in the CMAC is based on the various included outcomes in the MCDA and shows the probability of PC+ to be accepted as the preferred alternative (i.e. having the highest overall value), while the budget remains below a set threshold. This threshold refers to the budget available to be allocated to either PC+ or HBOC, for the treatment of a given population.

Moreover, we performed multiple imputation using multivariate imputation by chained equations (MICE), also known as fully conditional specification (FCS), to assess the impact of missing observations on the results of the CUA and MCDA [57]. Furthermore,

in an univariate sensitivity analysis, we used the Short-Form Health Survey with six dimensions (SF-6D) utilities to calculate QALYs instead of the EQ-5D-5L utilities used in the main analysis [58, 59]. This was done to make the results of the CUA more comparable to the results of the MCDA where quality of life was measured with the SF-12v2. Finally, local ranging standardisation of performance scores was used, instead of relative standardisation, to investigate the impact of the standardisation method on the MCDA results (details on this method are presented in Appendix 1).

RESULTS

In total, 1,783 patients from the intervention group and 272 from the control group with no missing observations in outcome and confounding variables were included. Before PSM, patients referred to PC+ had better physical health status and the distribution of the referred medical departments was uneven between the two groups (Table 2).

Selection of PSM technique and complete cases

Nearest Neighbor optimal matching with a 2:1 ratio was the best PSM technique, as it resulted in a high number of respondents with the least covariate imbalance (SMD › 0.25) and with acceptable Rubin's B (0.8%) and Rubin's R (0.984) (see Table S2). Following PSM, 544 patients from the intervention group and 272 from the control group were matched. However, in this matched sample, complete data on all outcome measures (i.e. criteria) during the three-month follow-up were available from 530 patients (65.0%), with 340 patients (62.5%) in the PC+ group, and 190 patients (69.9 %) in the HBOC group (see Table S3). As shown in Table 2, the differences in baseline characteristics between the two groups with complete cases were mitigated after PSM.

Table 2 Baseline Characteristics Before and After Propensity Score Matching

	Before PSM			
	PC+	**HBOC**	**P-value**	**SMD**
N	1783	272		
Age – mean (sd)	54.87 (15.96)	56.09 (16.00)	0.239	0.077
Gender (male) % (n)	38.6 (689)	34.9 (95)	0.268	0.077
Foreign-born % (n)	3.7 (66)	5.1 (14)	0.327	0.070
Low educational level % (n)	19.1 (340)	22.1 (60)	0.281	0.074
SF-12v2 PCS – mean (sd)	47.65 (9.26)	45.79 (9.94)	0.002*	0.194
SF-12v2 MCS – mean (sd)	50.99 (9.42)	50.13 (9.05)	0.158	0.093
Historical healthcare costs in € – mean (sd)	761.4 (2767.07)	811.7 (3069.00)	0.783	0.017
Medical specialty referred to			<0.001**	0.731*
Dermatology % (n)	31.8 (567)	12.1 (33)		
Gynaecology % (n)	5.8 (104)	9.6 (26)		
Internal medicine % (n)	2.5 (44)	9.2 (25)		
Otorhinolaryngology % (n)	17.0 (303)	12.5 (34)		
Neurology % (n)	7.7 (138)	4.8 (13)		
Ophthalmology % (n)	7.9 (141)	5.9 (16)		
Orthopaedics % (n)	19.3 (344)	39.7 (108)		
Rheumatology % (n)	6.6 (118)	3.7 (10)		
Urology % (n)	1.3 (24)	2.6 (7)		

Note: HBOC = Hospital Based Outpatient Care; MCS = mental component summary; PC+ = Primary Care Plus; PCS = physical component summary; PSM = Propensity score matching; SD = standard deviation; SF-12v2 = Short-Form Health Survey version 2; SMD = standardised mean differences;

** P < 0.01; ** P < 0.001; SMD > 0.25*

The illustrated graphics of the results of the PSM in terms of propensity score distributions and covariate balance are presented in Figure S1 and Figure S2. Regarding the doubly robust estimation using generalised linear regression models, the model performance based on the AIC and BIC is presented in Table S4 and Table S5.

Results of the CUA

The results of the CUA are presented in Table 3 and show that PC+ led to a cost reduction of €259.04 (95% CI = -447.03 - -71.05) and QALY gain of 0.002 (95% CI = -0.004 - 0.007), indicating therefore that PC+ was the dominant strategy.

	After PSM			
	PC+	P-value	SMD	
	340	190		
	58.03 (15.41)	59.80 (13.95)	0.190	0.121
	33.8 (115)	34.7 (66)	0.907	0.019
	5.9 (20)	5.3 (10)	0.920	0.027
	23.2 (79)	23.7 (45)	0.992	0.011
	45.22 (9.53)	45.10 (9.94)	0.891	0.012
	49.13 (10.16)	50.12 (9.15)	0.266	0.102
	772.22 (3239.36)	897.91 (3473.91)	0.677	0.037
			0.813	0.191
	7.9 (27)	11.1 (21)		
	9.7 (33)	8.4 (16)		
	8.2 (28)	7.9 (15)		
	11.5 (39)	15.3 (29)		
	4.7 (16)	4.2 (8)		
	5.9 (20)	6.8 (13)		
	44.7 (152)	40.5 (77)		
	3.8 (13)	3.7 (7)		
	3.5 (12)	2.1 (4)		

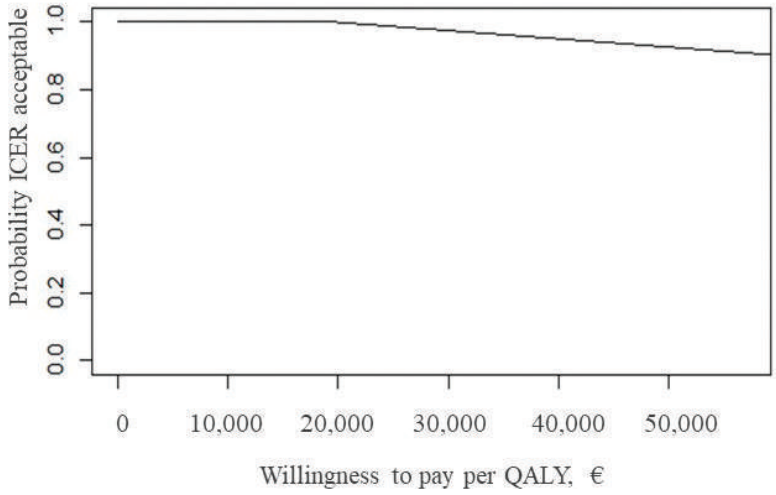
The cost-effectiveness (CE) plane with the bootstrapped ICERs (see Figure S3) shows that the majority of the bootstrapped ICERs (86.4%) are located in the south-east quadrant of the plane, indicating that PC+ is, besides less costly, also more effective compared to HBOC. However, a small proportion of points (13.6%) is located in the south-west quadrant, indicating that PC+ is less effective compared to HBOC. The CEAC (Figure 1) shows that the probability of PC+ being cost-effective decreases from 100% to 86.6% when the willingness to pay per QALY increases to €50,000, as a small proportion of bootstrapped ICERs was located in the south-west quadrant.

Table 3 Incremental cost-effectiveness ratio (ICER)

Alternative	Total cost (mean, sd)	Total QALY (mean, sd)	Incremental cost (95% CI)
PC+	490.34 (211.22)	0.191 (0.034)	-259.04(-447.03 – -71.05)
HBOC	763.54 (287.21)	0.188 (0.033)	

Note: SD = standard deviation; CI = Confidence Interval; QALY = quality-adjusted life-years; ICER = incremental cost-effectiveness ratio; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

[a] *Dominant = less costs, better outcomes*



Note: ICER = incremental cost-effectiveness ratio; QALY = quality-adjusted life-year

Figure 1 Cost-effectiveness acceptability curve

Incremental QALY (95% CI)	ICER
0.002 (-0.004 – 0.007)	Dominant [a]

Results of the MCDA

Aggregated performance for PC+ and HBOC is presented in Table 4. Psychological well-being was considered the most important criteria, with a stakeholders' mean weight of 0.308. Regarding the total scores, calculated by aggregating the average stakeholders' weights and standardised performance scores, the largest differences were found in the scores on accessibility (0.140 versus 0.126) and total costs (0.094 versus 0.061). In line with the result of the CUA, the overall total score shows that PC+ outperforms HBOC (0.730 versus 0.681). The unstandardised performance scores, the weights of the criteria from the viewpoint of the different stakeholders and the value scores with standardised performance based on relative scaling from these different perspectives are presented in Table S6, Table S7 and Table S8.

The CMAC (Figure 2) shows that the probability of PC+ being effective and affordable increases to 100% at a budget of €5,600,000 for the 3-month outpatient and inpatient hospital costs of 10,740 people eligible to be referred to PC+, or €521.42 per patient for the same period.

Table 4 Aggregated weights and standardised performance

Criteria	Standardised performance * score (se)	
	PC+	**HBOC**
Physical functioning	0.706(0.006)	0.708 (0.008)
Psychological well-being	0.710 (0.006)	0.704 (0.007)
Person-centeredness	0.707 (0.001)	0.708 (0.002)
Accessibility of care *	0.742 (0.004)	0.670 (0.006)
Total costs *	0.841 (0.020)	0.540 (0.015)
Overall total score		

Note: se = standard error; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

** Standardised performance based on relative standardisation with accessibility of care and total costs being reverse coded with a lower standardised score referring to a higher non-standardised score*

Stakeholders' mean weight (se)	Total score	
	PC+	HBOC
0.248 (0.013)	0.175	0.175
0.308 (0.011)	0.219	0.217
0.144 (0.007)	0.102	0.102
0.188 (0.013)	0.140	0.126
0.112 (0.014)	0.094	0.061
	0.730	0.681

Results of univariate sensitivity analyses

The results of the CUA and MCDA on the imputed dataset were similar to the results of the main analysis. However, in both analyses there was slightly more uncertainty regarding the effectiveness of PC+. This was illustrated by a higher number of bootstrapped ICERs (20.4%) in the southwest quadrant of the CE plane and a higher budget requested (€563.31 per patient) for PC+ to be effective and affordable on the CMAC (see Appendix 2). When SF-6D utility scores were used in the CUA instead of EQ-5D-5L utility scores, PC+ led to -0.000 (95% CI = -0.003 – 0.002) less QALYs, illustrated with a majority of bootstrapped ICERs (79.1%) in the southwest quadrant of the CE plane (see Appendix 3). Finally, although the overall value scores were lower when using local ranging standardisation instead of relative standardisation in the MCDA, PC+ was still the most preferred alternative (see Appendix 4).

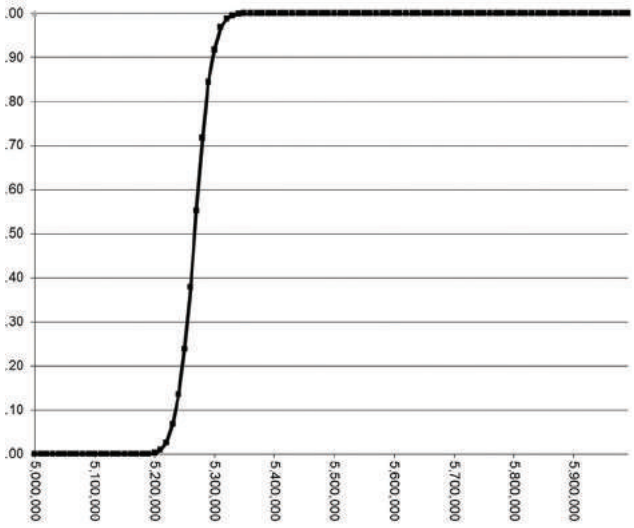


Figure 2 Conditional Multi-attribute Acceptability Curve

DISCUSSION

This study demonstrated a side-by-side performance of CUA and MCDA to evaluate a new model of care, PC+ for non-acute and low-complex patients, and found comparable results. Both methods of economic evaluation showed that PC+ was very likely to be cost-effective compared to HBOC. Although the decision suggested by the CUA and MCDA was the same, this study highlights a number of interesting dissimilarities and potential synergies between the two methods.

First, in the CUA the opportunity costs of one unit of additional benefit, in this case one additional QALY, is estimated. In the MCDA, a new composite score of benefit is created using the scores on all included criteria [26]. Therefore, opportunity costs need to be calculated determining one unit of additional benefit of that composite score. By including costs as a criterion, it is argued that the opportunity costs are not addressed adequately in the MCDA, and therefore, costs should not be included [60-63]. However, an argument in favour of including costs in the MCDA, just like the other criteria, is that the relative contribution of costs to the decision-making process is made more explicit [52]. Additionally, when implementing new models of care, reducing costs is one of the aims on new models of care, alongside improving quality of and access to care. Furthermore, if costs of a new model care reflect the value of the displaced interventions (e.g. care-as-usual), then opportunity cost is incorporated in the decision. Therefore, incorporating costs as a criterion in MCDA of new models of care may be useful in local decision-making.

Although CUA incorporates a multi-attribute measurement of outcome (i.e. longevity and quality of life), MCDA can incorporate many more outcomes and has the ability to decompose them to support multifactorial decisions. Such decisions may be applied more frequently because of the importance at local level where the decision is closer to the local needs and interests of several stakeholders. This is evident in the fact that local decision makers are using disaggregated outcomes (e.g. Key Performance Indicators) similar to a CCA [64]. In addition, MCDA can provide a systematic ranking of several alternative strategies that local decision makers may consider to optimise budget allocation [24]. However, multiple comparisons may be more complex in a CUA framework.

Furthermore, the CMAC seems to be a more suitable tool than CEAC to support local decision makers. This may be because it uses a wider range of outcomes, beyond the QALY, relevant for decision-making such as equity, patient satisfaction, and access

to care considerations, without requiring an additional axis and without increasing the complexity of the interpretability of the results. Additionally, the CMAC uses the budget available to be allocated across different models of care for a specific population instead of a range of willingness to pay thresholds for a QALY, which is less relevant to health and social care budget holders at local level.

Our study showed that the results of the CUA may be sensitive to utility measures that may even alter the decision. Using SF-6D utilities instead of EQ-5D-5L utilities changed the decision in our case study from dominant to cost-saving at lower QALYs. This change may be caused by the fact that the SF-6D is more efficient in detecting small changes in health-related quality of life [65]. The sensitivity of CUA results to the selection of the utility measure is evident in the literature [66] and may make CUA less attractive to local decision-makers as they often cannot determine the most appropriate utility measure to be routinely collected for the whole population in their catchment area. Additionally, the MCDA results showed little difference between PC+ and HBOC in the score on physical functioning and psychological well-being. The MCDA outcome of PC+ outperforming HBOC was mainly driven by accessibility of care and the total costs.

While MCDA is an exhaustive, flexible and inclusive approach with a growing popularity to evaluate interventions in healthcare, it also has some drawbacks [67, 68]. First, the subjectivity of the weighting approach in the MCDA, which is generally based on human judgment, can be subject to bias [68]. Together with the ad-hoc aggregation of outcomes, the results of the MCDA are context-dependent and therefore less generalizable to other situations. However, it should be mentioned that the QALY is also a preference-based health state classification system, using preferences from a general population sample [69]. Furthermore, it is questionable to what extent generalisability of results is desirable in decision-making in a local context. Although, it would be beneficial when local decision-makers set a core set of criteria and elicit the weights, which can be reused.

Additionally, compared to CUA, MCDA is often described as a complex and/or burdensome process [51, 70, 71]. This is related to some practical issues that might arise when using MCDA, such as the need to learn relevant techniques to perform MCDA or to have a facilitator to help using these techniques, the need to expand data collection to assess the full range of outcomes, the need for relevant software and/or programs to analyse data, and finally translate the model output in recommendations

[15, 72]. However, local decision makers are increasingly using decision support tools (e.g. balanced score cards and analysis of key performance indicators) with similar data and analytical needs with MCDA, which facilitate its feasibility and applicability at local level.

Furthermore, where the use of a CUA is straightforward, the MCDA is flexible to incorporate multiple objectives and to allow for the different goals between interventions and places. Therefore, the MCDA facilitates dialog and forces decision makers to think about and clearly express relevant values [73]. However, this flexibility is also challenging since many MCDA methods are available [67, 74]. The differences of these methods are not only related to practical issues, but also to the underlying fundamental theories and beliefs [75].

To overcome these practical challenges and to prevent the misuse of MCDA, specific attention for the use of the MCDA in the evaluation of new models of care in a local context is needed [75, 76]. Therefore, more interaction between researchers and decision makers is needed to deliberate on best approaches. Furthermore, MCDA can be of added value to the widely used CUA. With this, informed allocation of limited resources to improve local population outcomes and promote sustainability of local healthcare can be ensured.

LIMITATIONS

Regarding the case study used in this research, some limitations exist related to the design of the study, like the absence of costs related to primary care, the relatively short follow-up period of three-months and the small study sample (especially the control group). Therefore, comparing CUA and MCDA using other case studies without these limitations could be beneficial. However, this case study incorporated all necessary methodological steps, and therefore it serves as a valuable example. Moreover, in local decision-making, routinely collected data and non-randomisation is common and therefore, other case studies will almost certainly face similar limitations.

Furthermore, we performed PSM to reduce observed confounding. However, because of the limited time points of measurement available, it was not possible to adjust for unobserved confounding (e.g. by performing a difference-in-difference analysis). We also acknowledge that potential unobserved bias may have been transferred to

the imputed data. Although, we expect that unobserved confounding to be minimal as this was a relatively healthy population and the results were adjusted for many observed confounders.

In addition, we used the criteria weights of the SELFIE study, which included people with more complex healthcare needs (i.e. people with multi-morbidity) than the population included in our case study. Although this was not ideal, we expect that it had little impact on the total performance scores in the MCDA as a) the criteria in the SELFIE study were defined in terms of general outcome concepts grouped by the Triple Aim, which were very similar to our criteria and allowed us to use slightly different indicators to measure same outcomes as in SELFIE, and b) the SELFIE criteria weights are likely to be generalizable to national level as they were derived from approximately 750 Dutch stakeholders including patients, partners of patients, providers, payers, and policy makers from across the Netherlands [77].

Finally, as mentioned before, different MCDA methods which are based on different theories can be used in the evaluation of new models of care. In this case study, the value-based MAUT method was used [52]. Besides different value-based methods, other categories of MCDA methods are the outranking, reference level and goal programming methods. Using different MCDA methods may lead to different decision outcomes. However, regardless of the method used, the main goal of the MCDA is to structure the decision and to support the assessment process.

CONCLUSIONS

Regardless of the decision outcome, the MCDA is of added value to the CUA in the evaluation of new models of care as it allows the incorporation of relevant outcomes and perspectives of involved stakeholders in local decision-making. This may result in a more informative, auditable and transparent decision-making process at local level. However, because of the practical challenges related to the use of the MCDA approach, the question is to what extent it will be embedded in the existing local decision-making process. Therefore, further formalisation and validation of the MCDA approach in the evaluation of new models of care in a local context is required.

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ADDITIONAL FILES

Table S1 Description of indicators

Indicators	Description
EuroQol five-dimensional questionnaire with five levels (EQ-5D-5L)	The EQ-5D-5L is a widely used generic measure consisting of five questions on mobility, self-care, pain/discomfort, usual activities and anxiety/depression. Respondents choose from five response levels for each of the dimensions. A health state index score, ranging from -0.446-1 (worst- to best imaginable health status) can be calculated from individual health profiles using a country specific utility tariff.
Short-Form Health Survey version 2 (SF-12v2)	The SF-12v2 consists of 12 questions measuring the health status of respondents by means of two summary scores; a physical component summary (PCS) and a mental component summary (MCS). PCS and MCS scores from 0 (lowest level of health) to 100 (highest level of health) and can be obtained using the instrument's developers standard scoring algorithm.
Consumer Quality (CQ) index	Patient centeredness and accessibility of care are included in the CQ-index, which is a standardised method for measuring experiences of patients with healthcare. In this study, items derived from the Dutch CQ-index general practice and hospital outpatient care were used. Items scores ranged from 1-4. However, travel time was measured on a continuous scale with travel time in minutes.

Table S2 Results of matching techniques on covariance balance

Matching Technique	Intervention group (n)	Control group (n)
Unmatched	1783	272
Exact matching	0	0
Nearest Neighbor: greedy	272	272
Nearest Neighbor: greedy with ratio 2:1	544	272
Nearest Neighbor: caliper (0.25)	272	272
Nearest Neighbor: caliper (0.25) with ratio 2:1	542	272
Nearest Neighbor: caliper (0.25) with ratio 3:1	782	272
Nearest Neighbor: caliper (0.25) with replacement	1783	262
Nearest Neighbor: optimal	272	272
Nearest Neighbor: optimal with ratio 2:1	544	272
Full matching: with min. 1 and max. 2 matches	419	272
Genetic matching	1783	245

Note: SMD = Standardised Mean Difference (or Standardised Bias)

Covariates with SMD > 0.25 (n)	Maximum SMD	Rubin's B	Rubin's R
1	0.731	73.3%	0.914
NA	NA	NA	NA
7	3.806	159.9%	0.014
0	0.091	1.2%	0.974
2	0.445	23.1%	1.226
0	0.137	3.6%	0.968
0	0.178	10.3%	0.930
1	0.703	70.4%	0.904
0	0.155	0.03%	1.000
0	0.098	0.8%	0.984
0	0.206	16.8%	1.012
1	0.650	65.4%	0.911

Appendix 1 Formulas for standardisation

Formula for Relative standardisation

$$S_{aj} = \frac{x_{aj}}{\left(x_{aj}^2 + x_{bj}^2\right)^{1/2}} \qquad S_{bj} = \frac{x_{bj}}{\left(x_{bj}^2 + x_{aj}^2\right)^{1/2}}$$

S_{aj} = standardisation of the performance score (x) of alternative a (PC+) on criteria j relative to the combined performance score (x) of both alternatives a (PC+) and b (HBOC) on criteria j.

S_{bj} = standardisation of the performance score (x) of alternative b (HBOC) on criteria j relative to the combined performance score (x) of both alternatives a (PC+) and b (HBOC) on criteria j.

x = unstandardised performance scores
a = alternative a (in this case PC+)
b = alternative b (in this case HBOC)
j = criteria j

Whereby for reverse coded criteria, which should attain a lower, standardised score the higher the non-standardised score (in this case accessibility and costs), the additional formula is used:

$$S_{aj} = \frac{1}{S}$$

Formula for relative standardisation

The formula used for local scaling, with the worst and best performance scores taken as the observed range.

$$S_{aj} = \frac{X_{aj} - X_{min}}{X_{maz} - X_{min}}$$

Saj = standardisation of the performance score (x) of alternative a (PC+) on criteria j on the observed scale.

x = unstandardised performance scores
a = alternative a (in this case PC+)
min = minimum observed performance score
max = maximum observed performance score

Table S3 Complete cases per outcome measure after propensity score matching

	PC+	HBOC	Total
Total N	544	272	816
EQ-5D-5L utility score (%, N)	64.9 (353)	62.1 (169)	64.0 (522)
SF-12v2 physical score (%, N)	59.9 (326)	72.1 (196)	64.0 (522)
SF-12v2 mental score (%, N)	65.6 (357)	72.1 (196)	67.8 (553)
Person centredness (%, N)	80.9 (440)	82.0 (229)	82.0 (669)
Accessibility (%, N)	81.6 (444)	83.8 (228)	82.4 (672)
Costs (%, N)	100.0 (544)	100.0 (272)	100.0 (816)
Total for CUA (%, N)	64.9 (353)	62.1 (169)	64.0 (522)
Total for MCDA (%, N)	63.2 (344)	70.2 (191)	65.6 (535)
Total for CUA and MCDA (%, N)	62.5 (340)	69.9 (190)	65.0 (530)

Note: PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; EQ-5D-5L = EuroQol five-dimensional questionnaire with five levels; SF-12v2 = Short-Form Health Survey version 2; CUA = Cost Utility Analysis; MCDA = Multi-Criteria Decision Analysis

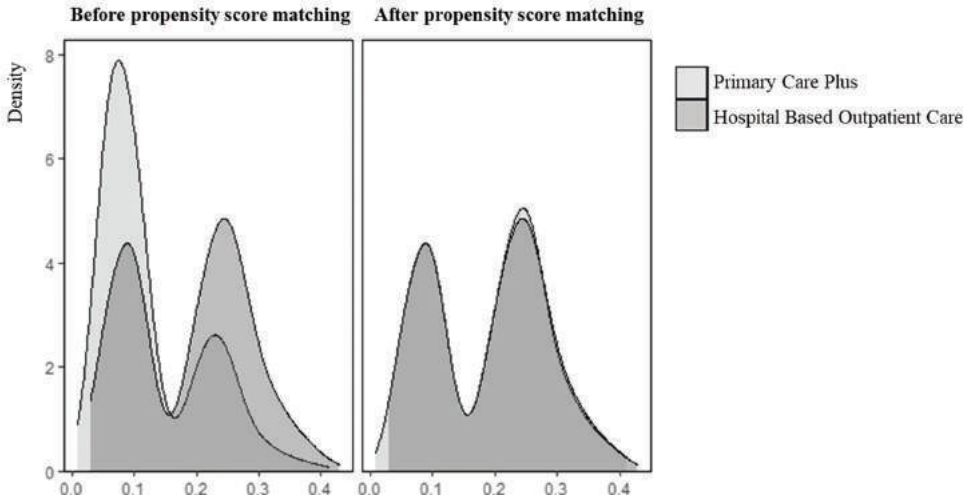


Figure S1 Propensity score distributions by study group before and after propensity score matching

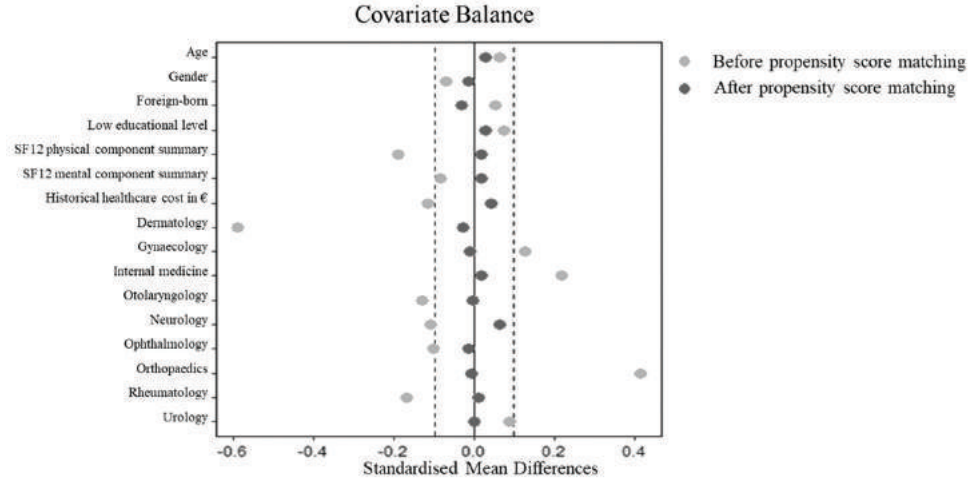


Figure S2 Standardised mean difference (or bias) before and after propensity score matching

Table S4 Estimating model performance for the Cost-Utility Analysis using the Akaike's and Bayesian Information Criteria

Distribution family	Link function	AIC	BIC
Cost			
Gamma	Log	6235.18	6282.18
Inverse-Gaussian	1/mu^2	n/a	n/a
Gaussian	Identity	9013.54	9060.55
QALYs			
Gamma	Log	-1867.93	-1816.65
Inverse-Gaussian	1/mu^2	-1479.57	-1428.29
Gaussian	Identity	-2974.97	-2623.69

Note: AIC = Akaike's Information Criterion; BIC = Bayesian Information Criterion; QALY = quality-adjusted life-year

Table S5 Estimating model performance for the Multi-Criteria Decision Analysis using the Akaike's and Bayesian Information Criteria

Distribution family	Link function	AIC	BIC
SF-12 physical score			
Gamma	Log	2841.49	2888.49
Inverse-Gaussian	1/mu^2	2938.31	2985.32
Gaussian	Identity	3420.16	3467.16
SF-12 mental score			
Gamma	Log	3010.93	3057.94
Inverse-Gaussian	1/mu^2	3123.15	3170.15
Gaussian	Identity	3603.92	3650.93
Person centredness			
Gamma	Log	746.77	793.77
Inverse-Gaussian	1/mu^2	933.46	980.46
Gaussian	Identity	674.05	721.05
Accessibility			
Gamma	Log	3467.52	3514.52
Inverse-Gaussian	1/mu^2	3450.79	3497.79
Gaussian	Identity	4453.39	4500.39
Cost			
Gamma	Log	6235.18	6282.18
Inverse-Gaussian	1/mu^2	n/a	n/a
Gaussian	Identity	9013.54	9060.55

Note: AIC = Akaike's Information Criterion; BIC = Bayesian Information Criterion

Table S6 Unstandardised performance scores after propensity score matching

Outcome measure	Unstandardised performance scores	
	PC+	HBOC
SF-12 physical score (mean, se)	46.31 (0.393)	46.46 (0.539)
SF-12 mental score (mean, se)	50.24 (0.395)	49.81 (0.468)
Person centredness (mean, se)	2.73 (0.006)	2.73 (0.007)
Accessibility (mean, se)	30.92 (0.186)	34.3 (0.299)
Costs (mean, se)	490.34 (11.45)	763.5 (20.84)

Note: PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SE = standard error

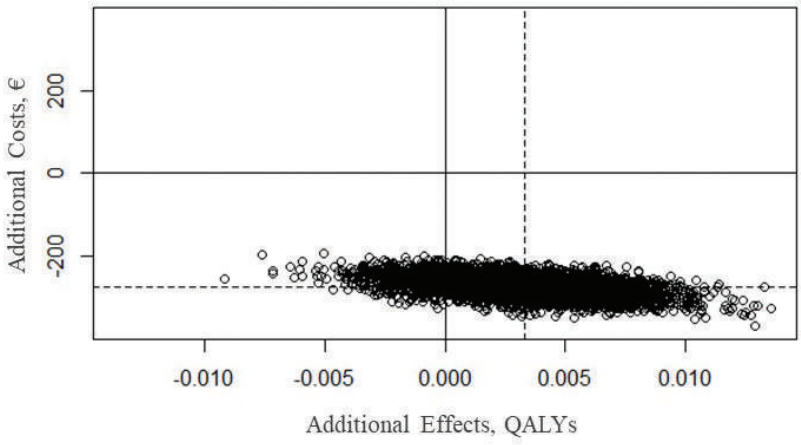
Table S7 The Dutch relative weights resulting from the SELFIE study

Criteria	Patients	Partners
Physical functioning	0.29	0.22
Psychological well-being	0.31	0.28
Person-centeredness	0.14	0.16
Continuity of care (i.e. accessibility)	0.19	0.23
Total costs	0.06	0.11

Table S8 Overall value scores with standardised performance based on relative scaling

Criteria	Patients		Partners	
	PC+	HBOC	PC+	HBOC
Physical functioning	0.205	0.205	0.155	0.156
Psychologicalwell-being	0.220	0.219	0.199	0.198
Person centeredness	0.099	0.099	0.113	0.113
Continuity of care (i.e. accessibility)	0.141	0.127	0.171	0.154
Total costs	0.050	0.032	0.093	0.059
Overall value score	0.715	0.682	0.731	0.680

Note: PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care



Note: QALYs = quality-adjusted life-years

Figure S3 Scatterplot of the estimated incremental costs and incremental effects of Primary Care Plus versus Hospital Based Outpatient Care obtained

Professionals	Payers	Policy makers
0.22	0.26	0.25
0.32	0.34	0.29
0.15	0.12	0.15
0.19	0.15	0.18
0.12	0.14	0.13

Professionals		Payers		Policy makers	
PC+	HBOC	PC+	HBOC	PC+	HBOC
0.155	0.156	0.183	0.184	0.176	0.177
0.227	0.226	0.241	0.240	0.206	0.205
0.106	0.106	0.085	0.085	0.106	0.106
0.141	0.127	0.111	0.100	0.134	0.121
0.101	0.065	0.118	0.076	0.109	0.070
0.730	0.680	0.738	0.685	0.731	0.679

Appendix 2 Sensitivity analysis using multiple imputation

Table 1 Baseline characteristics before and after propensity score matching when using multiple imputation

	Before PSM			
	PC+	HBOC	P-value	SMD
N	1831	285		
Age – mean (sd)	54.80 (15.99)	55.79 (16.08)	0.331	0.062
Gender (male) % (n)	38.5 (705)	35.1 (100)	0.299	0.071
Foreign-born % (n)	3.8 (69)	4.9 (14)	0.446	0.056
Low educational level % (n)	19.1 (349)	22.1 (63)	0.260	0.075
SF12 PCS – mean (sd)	47.68 (9.27)	45.81 (9.84)	0.002*	0.195
SF12 MCS – mean (sd)	50.95 (9.46)	50.17 (9.09)	0.193	0.084
Historical healthcare costs in € – mean (sd)	779.20 (2899.61)	569.82 (1972.51)	0.197	0.093
Medical specialty referred to			<0.001**	0.717*
Dermatology % (n)	31.7 (580)	12.3 (35)		
Gynaecology % (n)	5.7 (105)	9.5 (27)		
Internal medicine % (n)	2.6 (48)	8.8 (25)		
Otorhinolaryngology % (n)	17.0 (311)	12.6 (36)		
Neurology % (n)	7.7 (141)	5.3 (15)		
Ophthalmology % (n)	8.0 (146)	5.6 (16)		
Orthopaedics % (n)	19.3 (354)	39.6 (113)		
Rheumatology % (n)	6.6 (121)	3.5 (10)		
Urology % (n)	1.4 (25)	2.8 (8)		

Note: PSM = Propensity score matching; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care; SMD = standardised mean differences; SD = standard deviation; MCS = mental component summary; PCS = physical component summary

** P < 0.01; ** P < 0.001, * SMD > 0.25*

	After PSM			
	PC+	HBOC	P-value	SMD
	570	285		
	55.35 (16.25)	55.79 (16.08)	0.710	0.027
	35.8 (204)	35.1 (100)	0.899	0.015
	5.6 (32)	4.9 (14)	0.789	0.031
	20.9 (119)	22.1 (63)	0.745	0.030
	45.63 (9.67)	45.81 (9.84)	0.794	0.019
	50.03 (10.26)	50.17 (9.09)	0.840	0.015
	485.40 (1555.54)	569.82 (1972.51)	0.495	0.048
			0.997	0.076
	13.2 (75)	12.3 (35)		
	9.8 (56)	9.5 (27)		
	8.2 (47)	8.8 (25)		
	12.8 (73)	12.6 (36)		
	3.9 (22)	5.3 (15)		
	6.0 (34)	5.6 (16)		
	40.0 (228)	39.6 (113)		
	3.3 (19)	3.5 (10)		
	2.8 (16)	2.8 (8)		

Table 2 Incremental cost-effectiveness ratio (ICER) when using multiple imputation

Alternative	Total cost(mean, sd)	Total QALY(mean, sd)
PC+	537.42 (244.75)	0.184 (0.025)
HBOC	646.66 (325.24)	0.183 (0.024)

Note: SD = standard deviation; QALY = quality-adjusted life-year; CI = Confidence Interval; ICER = incremental cost-effectiveness ratio; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

[a] *Dominant = less costs, better outcomes*

Table 3 Aggregated weights and standardised performance when using multiple imputation

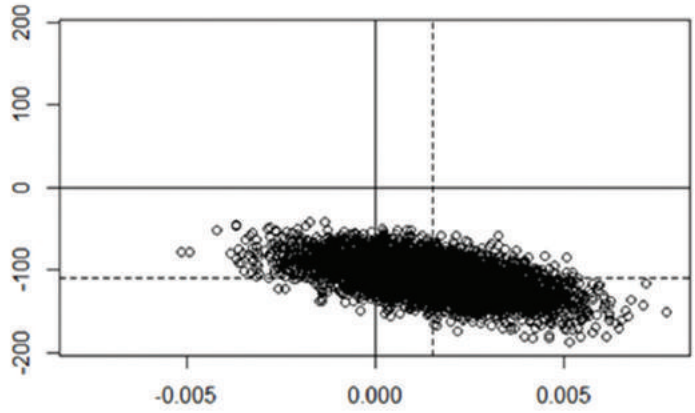
Criteria	Standardised performance * means (se)	
	PC+	HBOC
Physical functioning	0.707 (0.003)	0.707 (0.005)
Psychological well-being	0.709 (0.003)	0.705 (0.004)
Person-centeredness	0.713 (0.001)	0.701 (0.001)
Continuity of care (i.e. accessibility) *	0.757 (0.002)	0.653 (0.004)
Total costs *	0.769 (0.015)	0.639 (0.019)
Overall value score		

Note: se = standard error; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

** Standardised performance based on relative standardisation with continuity of care and total costs being reverse coded with a lower standardised score referring to a higher non-standardised score*

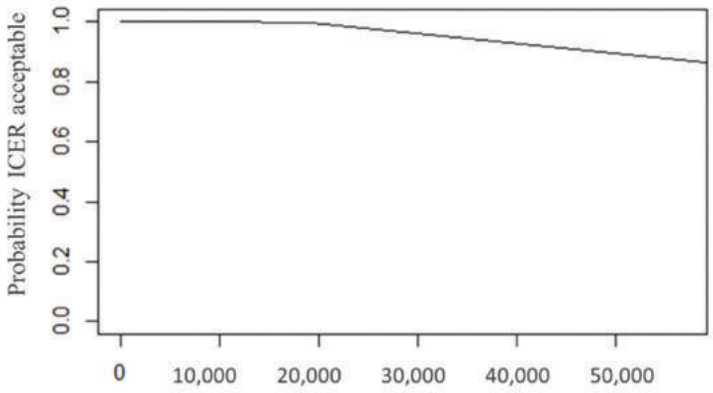
Incremental cost (95% CI)	Incremental QALY (95% CI)	ICER
-101.90 (-243.40 – 39.60)	0.001(-0.003 - 0.005)	Dominant [a]

Stakeholders' average weight (se)	Weighted aggregation	
	PC+	HBOC
0.248 (0.013)	0.175	0.175
0.308 (0.011)	0.218	0.218
0.144 (0.007)	0.103	0.101
0.188 (0.013)	0.142	0.123
0.112 (0.014)	0.086	0.072
	0.724	0.689



Note: QALYs = quality-adjusted life-years

Figure 1 Scatterplot of the estimated incremental costs and incremental effects of Primary Care Plus versus Hospital Based Outpatient Care obtained by bootstrap simulations when using multiple imputation



Note: ICER = incremental cost-effectiveness ratio; QALY = quality-adjusted life-year

Figure 2 Cost-effectiveness acceptability curve when using multiple imputation showing the probability that Primary Care Plus is cost-effective when compared with usual care over a range of willingness to pay for an additional quality-adjusted life-year (QALY)

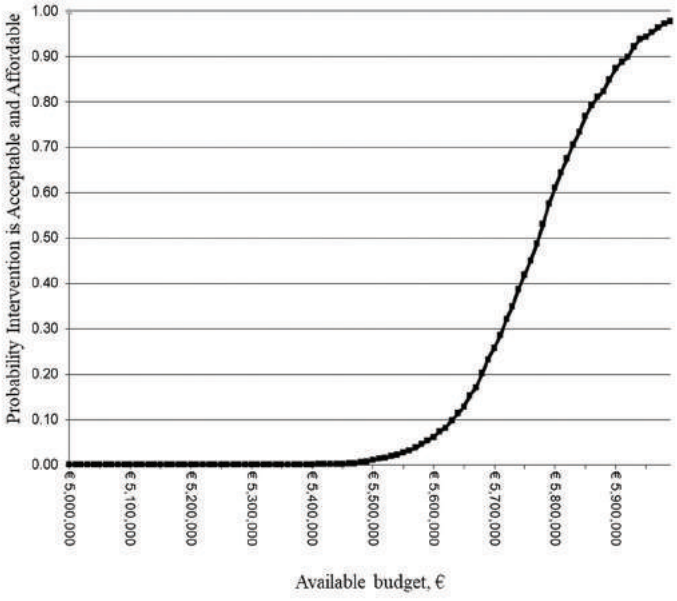


Figure 3 Conditional Multi-attribute Acceptability Curve when using multiple imputation showing the probability that Primary Care Plus is cost-effective when compared with usual care over a range of willingness to pay thresholds

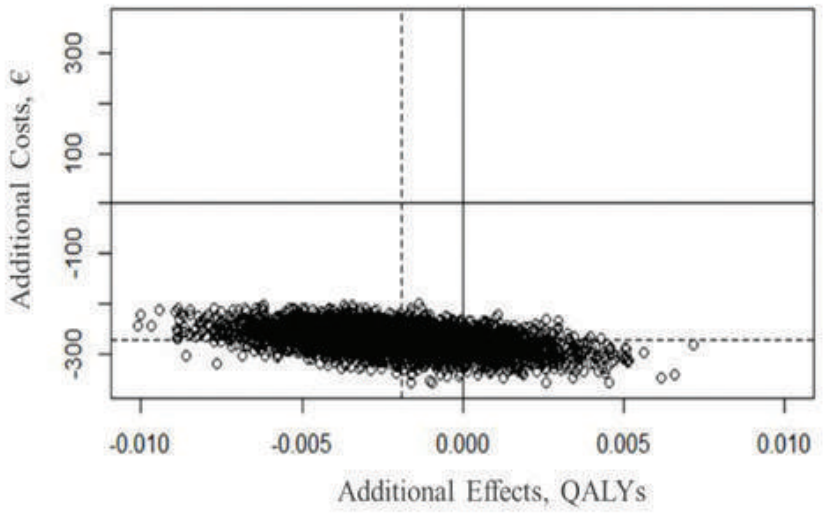
Appendix 3 Sensitivity analysis for Cost-Utility Analysis using the Short-Form Health Survey with six dimensions (SF-6D)

Table 1 Incremental cost-effectiveness ratio (ICER) when using the Short-Form Health Survey with six dimensions (SF-6D)

Alternative	Total cost (mean, sd)	Total QALY (mean, sd)	Incremental cost (95% CI)
PC+	490.34 (211.22)	0.163 (0.026)	-259.04 (-447.03 – -71.05)
HBOC	763.54 (287.21)	0.165 (0.027)	

Note: SD = standard deviation; CI = Confidence Interval; QALY = quality-adjusted life-years; ICER = incremental cost-effectiveness ratio; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

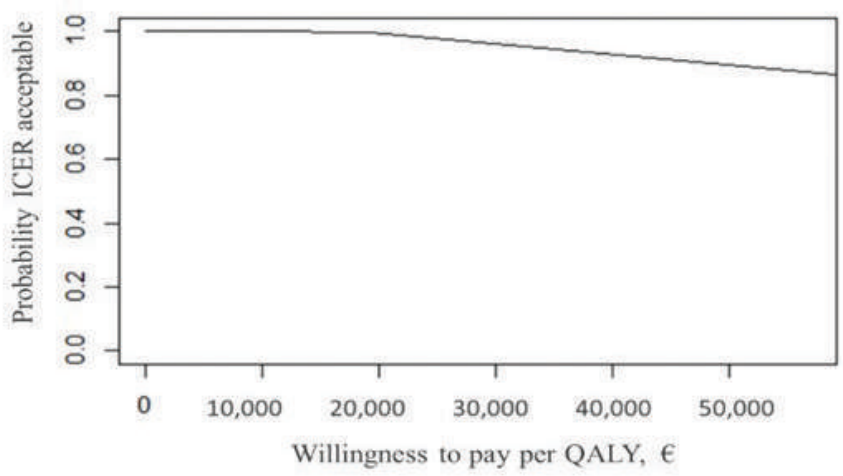
[a] *Cost-saving = less costs, worse outcomes*



Note: QALYs = quality-adjusted life-years

Figure 1 Scatterplot of the estimated incremental costs and incremental effects of Primary Care Plus versus Hospital Based Outpatient Care obtained by bootstrap simulations when using the Health Survey with six dimensions (SF-6D)

Incremental QALY (95% CI)	ICER
-0.000 (-0.003 – 0.002)	Cost-saving [a]



Note: ICER = incremental cost-effectiveness ratio; QALY = quality-adjusted life-year

Figure 2 Cost-effectiveness acceptability curve when using the Health Survey with six dimensions (SF-6D) showing the probability that Primary Care Plus is cost-effective when compared with care-as-usual over a range of willingness to pay for an additional quality-adjusted life-year (QALY)

Appendix 4 Sensitivity analysis for Multi-Criteria Decision Analysis using local ranging standardisation

Table 1 Aggregated weights and standardised performance when using local ranging standardisation

Criteria	Standardised performance * means (se)	
	PC+	HBOC
Physical functioning	0.483 (0.015)	0.479 (0.011)
Psychological well-being	0.590 (0.012)	0.577 (0.014)
Person-centeredness	0.557 (0.009)	0.563 (0.012)
Continuity of care (i.e. accessibility) *	0.787 (0.007)	0.663 (0.011)
Total costs *	0.900 (0.004)	0.811 (0.007)
Overall value score		

Note: se = standard error; PC+ = Primary Care Plus; HBOC = Hospital Based Outpatient Care

** Standardised performance based on local ranging standardisation with continuity of care and total costs being reverse coded with a lower standardised score referring to a higher non-standardised score*

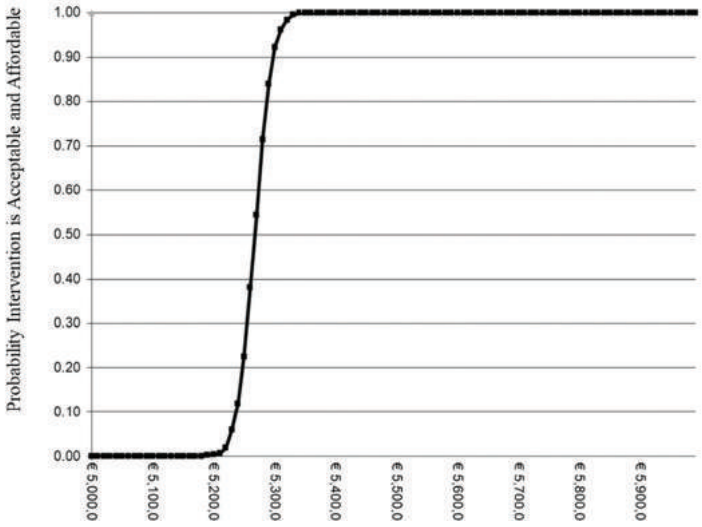


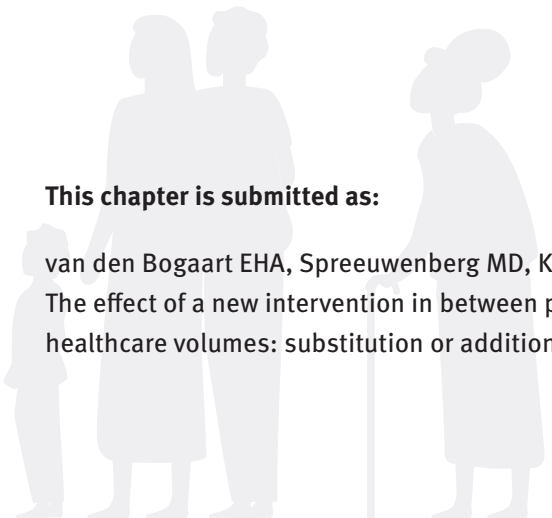
Figure 1 Conditional Multi-attribute Acceptability Curve when using local ranging standardisation showing the probability that Primary Care Plus is cost-effective when compared with care-as-usual over a range of available budgets

Stakeholders' average weight (se)	Weighted aggregation	
	PC+	HBOC
0.248 (0.013)	0.119	0.120
0.308 (0.011)	0.182	0.178
0.144 (0.007)	0.080	0.081
0.188 (0.013)	0.148	0.125
0.112 (0.014)	0.101	0.091
	0.630	0.595

07

Substitution or addition?

The effect of a new intervention in between primary and secondary care on regional healthcare volumes



This chapter is submitted as:

van den Bogaart EHA, Spreeuwenberg MD, Kroese, MEAL, Ruwaard D.
The effect of a new intervention in between primary and secondary care on regional healthcare volumes: substitution or addition?

ABSTRACT

To shift low-complex specialised care from the hospital to the primary care setting, the Dutch initiative Primary Care Plus (PC+) was implemented in 2014. Despite the positive effect of PC+ at the individual patient level concerning health-related quality of life, perceived quality of care and care costs, it is not clear whether and how PC+ affects regional healthcare volumes at the population level. Previous research describes possible mechanisms by which PC+ could be a substitution for or an addition to other types of health services. Therefore, this study used retrospective health insurance reimbursement claims data from the dominant health insurance company in the region to determine regional care volumes from 2015 to 2018 and compare them to the national trend. The results show an increase in the total volume of low-complex specialised care over time in the intervention region, by which PC+ did not fully succeed as a substitute for hospital services. Therefore, this study supports the idea that, in addition to measuring these kinds of interventions' effects on the individual patient level, the effects on the total healthcare utilization at a population level are necessary. However, the slightly decreasing volume trend at the end of the study period may indicate that substitution can be realised in the long term, which confirms that time is needed for these kinds of interventions to be effective. Thus, a longitudinal dynamic evaluation approach to inform policy makers is needed.

INTRODUCTION

How to control healthcare costs is a challenge in political debates worldwide [1]. Rising healthcare expenditures are threatening the financial sustainability of our healthcare systems [2]. Therefore, these systems are being challenged to deliver high-quality care at acceptable costs [3]. As a response to this need, there is an increasing interest in many healthcare systems to shift some type of care out of hospitals into the community [4, 5].

By shifting hospital care, an effort is being made to improve access to quality care and increase the effectiveness and efficiency of healthcare provision and delivery [6, 7]. In primary-care-based health systems, primary care acts as a gatekeeper and coordinator of care with a first-contact function that facilitates entry into the rest of the health system [8, 9]. As a result, primary care could have a major impact on the total costs of healthcare [10]. Therefore, various initiatives are being implemented to increase the emphasis on primary care to prevent, delay or avoid unnecessary and expensive hospital care [11]. Examples of shifting hospital care to the primary care setting are discharges from outpatient to primary care, shifted outpatient clinics, specialist attachment to primary care teams and joint consultations [12, 13].

In 2013, regional collaboration initiatives in the Netherlands were established to achieve 'better healthcare at lower cost' by focusing on the substitution of hospital care with primary care. One of these initiatives is Primary Care Plus (PC+), in which hospital specialists provide care in a primary care setting [14-16]. With this intermediate service, PC+ addresses low-complex and non-acute patients in a lower cost-per-unit setting to keep patients out of the hospital. Because PC+ care is provided by the same specialist as in the hospital but closer to patients' homes, it was hypothesised that the patient's health status and quality of care would not be affected. These assumptions were confirmed by previous research on PC+, showing that health and the quality of care were maintained or even improved [17, 18]. Moreover, previous research showed that with a lower cost-per-unit setting, achieved through lower overheads and less resource use, PC+ resulted in lower costs per patient compared to care-as-usual [17].

Despite these positive effects of PC+ on health-related quality of life, perceived quality of care and care costs at the individual patient level, it is not clear whether PC+ actually succeeds in shifting outpatient hospital care to the primary care setting and results in a substitution of care on a population (or regional) level. If PC+ appears

to be a substitution for (more expensive) hospital care, the positive effect on care costs at the patient level will be enhanced, and overall, PC+ will be a cost-saving initiative at a population level. However, if PC+ does not succeed in replacing hospital services but adds to them, it is unclear whether PC+ contributes to cost savings at the population level. Therefore, this study aims to investigate the effect of PC+ on regional healthcare volumes.

Mechanisms for substitution or addition

As described by Fortney et al. [19], a number of possible mechanisms exist by which primary care could be a substitution for or an addition to other types of health services. A promoting factor for the substitution mechanism of PC+ is the reinforcement of the gatekeeping role of primary care by embedding specialist knowledge into primary care [20]. With this knowledge, the prevention or early detection of illnesses can be achieved by offering more accessible specialised medical care [19]. As a result, patients can be treated in primary care, and referrals to hospital outpatient care can be prevented. Furthermore, prevention or delay of the need for hospital care can be achieved by focusing on patient education to promote self-management [19, 21]. Moreover, as medical specialists move closer to general practitioners (GPs), PC+ might encourage interaction and communication between healthcare professionals, resulting in opportunities for education [22]. In the long term, this might improve GPs' knowledge and skills and reduce the number of referrals to hospital care [23, 24].

On the other hand, there are factors that may enable PC+ to add to or supplement the existing volume of care. In this case, there is no or only limited substitution of hospital care with primary care; therefore, PC+ might have a negative effect on costs and capacity [25]. A factor that may explain this mechanism is the detection of illnesses that cannot be appropriately treated in PC+, and hence, a proportion of patients still need to be referred for hospital care following PC+ [14, 26, 27]. This mechanism may cause duplicate consultations and/or treatment, leading to additional costs. Furthermore, the ready availability of specialist knowledge in PC+ may increase the volume of care when GPs' referral thresholds are lowered. This is related to the theory of supply-induced demand, which may lead to an increase in the use of services [28].

METHODS

Setting

The PC+ initiative investigated in this study is located in the Maastricht-Heuvelland region, situated in the province of Limburg in the southern Netherlands. In this region, approximately 81 GPs working in 55 GP practices take care of a population of approximately 170,000 inhabitants [29]. VGZ is the health insurance company with the largest market share in this region and a PC+ stakeholder, along with the regional primary care organisation 'Care in Development' (in Dutch: Zorg In Ontwikkeling); the regional hospital Maastricht University Medical Centre + (Maastricht UMC+), with a combined function of an academic and a regional hospital; and the patient representative foundation 'Burgerkracht Limburg'. The partnership among the different organisations, and, in particular, between the primary care organisation and the hospital, is characterised by a long history of collaboration [30-33].

With the 2014 opening of two independent PC+ centres located outside the hospital premises, GPs within the region were able to refer patients with non-acute and low-complex health complaints to a medical specialist in a neutral primary care setting. The referral process for PC+ was similar to referring patients to outpatient hospital care. Since almost every Dutch resident is registered at a GP practice, the GP acts as a gatekeeper by referring patients to a medical specialist if needed [34]. Based on various criteria, such as patients' and GPs' preferences, familiarity with PC+, a patient's medical history, and wait and travel times, GPs decide in consultation with the patient whether to refer them to PC+. In PC+, patients are examined and/or treated by a (senior) medical specialist. These medical specialists are employed in the regional hospital and perform PC+ consultations on a regular basis. Since the hospital in the Maastricht-Heuvelland region is an academic hospital, all medical specialists are salaried [35]. After one or two consultations in PC+, patients are referred back to their GP with treatment advice. However, the medical specialist can also advise in referring the patient to hospital care when further diagnosis and/or treatment was needed. Previous research comparing PC+ with care-as-usual showed that in PC+, health and quality of care were maintained or even improved and PC+ resulted in lower costs per patient [17, 18].

Between January 2015 and December 2018 22,136 patients visited PC+ for one of the eleven different medical specialties (see Table 1). The composition of medical specialties present in PC+ changed during the study period. Additionally, there was

a difference in the number of PC+ consultations per medical specialty caused by the availability of personnel and the variation in GP referrals. Medical specialties that have been present from the start of PC+ started in November 2014.

Table 1 Patients visiting Primary Care Plus for the different medical specialties from January 2015 to December 2018 (N = 22,136)

Medical specialty	Number of patients % (N)	Start in PC+ *	Drop out
Dermatology	26.1% (5780)	Present from the start	
Orthopaedics	16.1% (3563)	Present from the start	
Internal medicine (including gastroenterology)	3.5% (770)	Present from the start	
Neurology	5.8% (1290)	Present from the start	
Ophthalmology	7.5% (1661)	Present from the start	
Rheumatology	7.0% (1545)	Present from the start	
Otorhinolaryngology	16.1% (3556)	January 2015	
Paediatrics	0.2% (50)	December 2015	January 2017
Gynaecology	11.9% (2633)	December 2015	
Urology	1.1% (238)	March 2016	November 2017
Back pain consultation facility	4.7% (1050)	November 2016	

Note: PC+ = Primary Care Plus

** Medical specialties that have been present from the start of PC+ in November 2014*

The Dutch Healthcare System

As PC+ is located in the primary care setting, consultations are fully covered by health insurance, and no compulsory deductible is levied for a consultation with a medical specialist in PC+. This exemption is a benefit to the patient because when consulting a medical specialist in outpatient hospital care, patients have to pay a compulsory deductible up to a maximum amount [36]. This compulsory deductible is annually determined by the Dutch government, and insured persons have the option to increase this amount in return for a premium discount [37, 38]. During the study period, the compulsory annual deductible ranged from €375 to €385.

Furthermore, PC+ is reimbursed on a fee-for-service basis with a fixed price per consultation, regardless of the medical specialty. This differs from the reimbursement of hospital care, for which a detailed hospital product classification system has been used since 2005, in which each patient is categorised into a Diagnosis Treatment Combination (DTC) [39]. These DTCs include all hospital activities and services (both

inpatient and outpatient) associated with the patient's demand for care from the initial to the final consultation or examination. Hospitals receive a payment per DTC that has been agreed upon with the insurer.

Data and analyses

In this observational study, retrospective health insurance reimbursement claims data from the dominant health insurance company (VGZ) in the Maastricht-Heuvelland region were used to determine the volume of care. With these data, PC+ and secondary care volumes (i.e. volumes of the regional hospital and volumes of other secondary care settings in and outside the region visited by patients from the intervention region) from 2015 to 2018 were described and compared to the national trend. Despite that the PC+ centres opened in 2014, this year is not included in the data due to a different method of invoicement than in the following years. In addition, the PC+ volume was still very low in 2014 as the start-up was only in November. Because claims data are available with a delay, extending the follow-up period after 2018 was not possible.

Claims were selected when the referring physician was a GP within the intervention region. Regarding the claims related to secondary care, a selection was made for low-complex initial hospital care (usually involving one or two consultations with a medical specialist) suitable for substitution to PC+ based on the DTC system (see Appendix I). Only DTCs related to a medical specialty present in PC+ during the study period were selected (see Table 1). The annual number of claims was adjusted for the number of registered insured persons by the number of claims per 1,000 insured. To compare the volume of care in the intervention region with the Dutch average over time, a comparable data selection was made for claims in the rest of the Netherlands related to VGZ-insured persons. The intervention region was excluded from this selection.

Additionally, data from the primary care organisation in the intervention region and Statistics Netherlands (in Dutch: Centraal Bureau voor de Statistiek) were used to calculate the annual number of inhabitants in the intervention region and the rest of the Netherlands. These numbers have been used to calculate the annual percentage of VGZ-insured persons.

This study was approved by the Medical Research and Ethics Committee of Maastricht University Medical Centre (METC 14-4-136).

RESULTS

Table 2 shows the total population and the number of VGZ-insured persons from 2015 to 2018, both in the intervention region and in the rest of the Netherlands. During the study period, the population averaged 169,125 inhabitants in the intervention region. On average, 46.5% of this population was VGZ-insured. In the rest of the Netherlands, approximately 21.0% of the average 16,866,484 inhabitants were VGZ-insured during this period.

Furthermore, Table 2 shows the number of claims related to low-complex specialised care to describe the volume of care over time in the intervention region (regional level) and in the rest of the Netherlands (national level). Within the intervention region, a distinction was made between care provided in PC+; in the regional hospital; and in other secondary care settings, such as independent treatment centres and other hospitals outside the intervention region. The total number of claims per 1,000 insured related to low-complex specialised care in the intervention region rose from 167.5 in 2015 to 184.2 in 2018. The increase in claims was related to PC+, which rose from 24.7 claims per 1,000 insured in 2015 to 48.8 in 2018. The number of claims related to the regional hospital decreased over time, from 89.7 claims per 1,000 insured in 2015 to 82.9 claims in 2018. The number of claims in other secondary care settings decreased from 53.1 claims per 1,000 insured in 2015 to 52.5 claims in 2018. At the national level, the total number of claims decreased from 141.1 claims per 1,000 insured in 2015 to 135.5 in 2018.

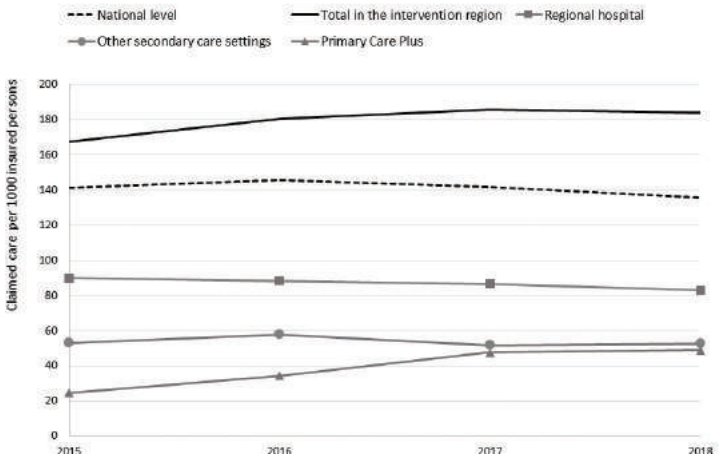


Figure 1 Trends in low-complex specialised care claims per 1,000 VGZ-insured persons from 2015 to 2018

In Figure 1, the volume trend per 1,000 insured persons over time in the intervention region and at the national level are presented. It shows that the volume of care in PC+ is, as expected, increasing over time. Because the decreasing trend in the volume of low-complex specialised care in the regional hospital is limited, an increasing trend in the total volume of care in the intervention region over time is visible. During the same period, a declining trend at the national level is observed. Furthermore, a volume difference between the intervention region and the national level over the whole study period is visible, with a higher total volume of low-complex specialised care in the intervention region.

Table 2 Number of (insured) persons and low-complex specialised care claims from 2015 to 2018 at the regional and national levels

Regional level	**2015**
Regional population (N)	168,660
VGZ-insured persons in the intervention region % (N)	45.3% (75,821)
Claims per 1,000 insured (total N) [A]	
In Primary Care Plus	24.7(1,873)
In the regional hospital	89.7 (6,801)
In other secondary care settings	53.1 (4,024)
In total	167.5 (12,698)
National level	**2015**
National population (N) [B]	16,732,066
VGZ-insured persons in the rest of the Netherlands % (N)	20.7% (3,464,402)
Claims per 1,000 insured (total N) [A]	
In total	141.4 (489,864)

[A] *Claims are related to low-complex specialised care for a selection of medical specialties;*

[B] *National population minus the intervention region (data retrieved from Central Bureau for Statistics [40])*

2016	2017	2018
170,385	167,052	170,403
46.9% (79,325)	47.8% (79,264)	46.1% (78,503)
34.4 (2,727)	47.5 (3,767)	48.8 (3,829)
88.3 (7,008)	86.6 (6,862)	82.9 (6,510)
57.8 (4,585)	51.7 (4,100)	52.5 (4,120)
180.5 (14,320)	185.8 (14,729)	184.2 (14,459)

2016	2017	2018
16,808,735	16,914,455	17,010,681
20.5% (3,451,320)	21.1% (3,576,861)	21.5%(3,656,494)
145.7 (502,734)	141.6 (506,642)	135.5 (495,591)

DISCUSSION

Previous research has shown that PC+ is able to maintain health and the quality of care at lower costs at the individual patient level [17, 18]. In addition to these findings, this study explored the effect of PC+ on the regional healthcare volume to investigate whether PC+ indeed results in a substitution of care at the population level. This study's results show an increase in the total volume of low-complex specialised care in the intervention region over time, which was not found at the national level. Despite the decreasing trend in the regional hospital volume of care, complete substitution of care was not achieved. Therefore, during the study period, PC+ has not been able to fully substitute for hospital care, and consequently, the volume of care of PC+ appears to be partly an addition to the volume of care in secondary care.

As mentioned in previous research, a factor that may enable PC+ to add partly to the existing volume of care is related to the detection of illnesses in PC+, necessitating a referral to hospital care [14, 26, 27]. However, previous research shows that approximately one-fifth of patients received advice to be referred to secondary care for further diagnosis and/or treatment following their PC+ consultation [14, 26]. Therefore, this mechanism appears to be less relevant in this situation. Another mechanism mentioned in the literature is related to supply-induced demand by which the implementation of PC+ causes an increase in the use of healthcare services [28]. This could be the case when PC+ lowers the threshold for GPs to refer to specialised care, resulting in more (unnecessary) referrals [41]. On the other hand, supply-induced demand can also be related to the response of secondary care to a possible volume reduction. In this situation, medical specialists may increase the demand for services by recommending procedures and services that would not otherwise be conducted [42] by requesting that patients visit the hospital more frequently [43], or by attracting new and different patient groups [44]. This phenomenon in which the compression in one part of the system leads to expansion elsewhere is referred to as the 'balloon' effect [45]. However, it is questionable whether this mechanism is relevant in this situation since all specialists involved in this study were salaried. Finally, supply-induced demand can also be related to patients' utilisation of medical care. This can be referred to as the effect of moral hazard: individuals, on average, reduce their (unnecessary) healthcare consumption when financial consequences are involved [46, 47]. Because PC+ is excluded from the compulsory deductible, the effect of moral hazard can be diminished.

The volume difference between the intervention region and the rest of the Netherlands during the study period can be explained by the health disadvantage in the intervention region [48]. This disadvantage arose because the province of Limburg is characterised by, among other things, lower labour participation, an unhealthy lifestyle, and more educational disadvantages. As a result, the volume of healthcare utilisation in Limburg is higher than elsewhere in the Netherlands. However, due to the lower level of the gross domestic product per capita and a lack of highly skilled workers, this region might also be affected by cost-related healthcare access problems [49]. As a result, patients' demand for care may have increased because no compulsory deductible is levied for a consultation with a medical specialist in PC+. According to this mechanism, PC+ could have contributed to the accessibility and timeliness of specialised care in the intervention region, whereby care is provided with minimal delays and barriers [50]. By improving access, the timeliness of health services can be increased to achieve the best possible health outcome [51]. However, the data used in this study do not provide further insights into the possible mechanisms that can explain the different volume trends. Therefore, based on these data, further (qualitative) research is needed to understand what is actually occurring in the region. With this information, policy can be created that focuses on influencing certain mechanisms to achieve substitution.

Moreover, to eliminate the difference in population characteristics between the intervention region and the rest of the Netherlands, adjustments should be made for characteristics such as age and gender. By correcting for these characteristics, the probability of patients' utilisation of medical care is taken into account. Because the trend analyses in this study are explorative in nature, such corrections have not been made. In addition, the question is whether the limited decline in the volume of low-complex specialised care that is ultimately visible in the intervention region can actually be attributed to substitution since an even stronger declining trend at the national level is observed during the same period. However, it should be noted that in the rest of the Netherlands no substitution initiatives have been included in the claims data. Because registration of these types of initiatives is ambiguous, they are difficult to trace from health insurance reimbursement claims data. By excluding initiatives such as PC+, the volume of low-complex specialised care in the rest of the Netherlands may be underestimated. In fact, without the PC+ volume, the intervention region would also show a decreasing trend over time. Finally, in addition to volume, total costs should also be considered. Even though the volume of low-

complex specialised care increased in the intervention region, the lower costs per patient in PC+ can still lead to eventual cost savings on a population level.

This study supports the idea that, in addition to evaluating the outcomes of interventions, such as PC+, at the individual patient level, it is also important to evaluate the outcomes at the population level, i.e., at the patient level, regionally and nationally. Continuously evaluating and monitoring the intervention at both the individual and population levels ensures that policy makers are informed about service development and, if necessary, possible adjustments can be made [44, 52]. Additionally, according to Fulop et al. [53], there is a tendency to underestimate the time needed for integrated care interventions to be effective. Implementing new models of care, such as PC+, requires a complex cultural and organisational change to deliver care differently [54]. Therefore, a longitudinal dynamic evaluation over a longer period of time to assess the impact on different outcomes, such as costs of care, healthcare consumption, and health-related outcomes, is needed [55]. The relatively short follow-up period in this study may result in the risk of concluding prematurely that PC+ did not result in a substitution of care [56]. However, a slightly decreasing trend in the volume of care in the intervention region is visible in 2018, which may show that substitution can eventually be achieved. Though policymakers should realise that with these kinds of initiatives costs go before earnings, and therefore, stakeholder courage is needed to implement new models of care.

Generally, as stated by Fries et al. [57], broadening access to care and reducing the overall volume, and thereby costs of care, at the same time is difficult. Therefore, to achieve the joint objective of providing "better healthcare at lower costs", more emphasis needs to be given to incentives and/or payment systems aligned with this objective [58, 59]. For example, in the Bernhoven case, a contract innovation for healthcare reimbursement consisting of a 5-year contract runtime instead of a year-to-year construction was implemented [58]. This multi-year contract guarantees stable income for the hospital, regardless of the volume and burden of claims, and ultimately contributes to a cultural shift towards a more quality-driven system. Another well-known example is Gesundes Kinzigtal in Germany, in which a long-term shared savings contract with health insurers was implemented [60]. Therefore, shifting to payment models more aligned with the Triple Aim [61], such as bundled or global payments, may increase financial accountability for healthcare providers. An example is the global budget contract implemented in the Alternative Quality Contract (AQC) in the U.S. [62]. Based on a global budget and pay-for-performance for achieving certain

quality benchmarks, this contract puts providers at risk for excessive expenditure and rewards them for quality. This type of (financial) incentive seems necessary because, according to Shortell et al. [63], only convincing healthcare providers of the clinical argument to change service delivery is often insufficient.

LIMITATIONS

Though health insurance reimbursement claims data are a valuable research resource, they also have limitations because these data were not originally acquired for research purposes [64]. In this study, limitations were experienced related to the fact that the medical specialty referred to was not traceable from the PC+ claims data. Therefore, it was not possible to investigate the volume developments per medical specialty and to take into account the in- and outflow of the different medical specialties. Because each medical specialty has a different share in PC+ and a different approach, it is expected that there will be different effects on the volumes of care in both PC+ and secondary care. The option to link the data with other resources to provide insights into the medical specialty referred to was not possible due to privacy issues.

Furthermore, only claims data from PC+ and secondary care were included to study the volume trends over time. However, including claims data related to primary care might also be useful, since a majority of the patients referred to PC+ are again referred to their GP following PC+. Therefore, PC+ can also affect the healthcare volume in primary care, and the increasing volume effect caused by PC+ may be underestimated in this study. However, the cost of primary care is substantially lower than that of secondary care.

Finally, only claims data from health insurance company VGZ were included. Although this health insurance company has the largest market share in the intervention region, only including the VGZ population may cause selection bias and limit the external validity of the results [65]. For example, it is not clear to what extent the study population is representative of the total population in the intervention region. The same applies to the population in the rest of the Netherlands, where the share of VGZ-insured persons is even lower than in the intervention region. Using claims data from several health insurance companies could increase generalisability. However, linking multiple data sources can be a challenge since data can be defined differently [64].

CONCLUSIONS

Although PC+ results in positive outcomes at the individual patient level regarding health-related quality of life, perceived quality of care and care costs, this study shows that the desired results related to the volume of low-complex specialised care at a population level are limited. Due to the limited decrease in the volume of care in the regional hospital, PC+ has an additional effect on the total volume of care on a population (or regional) level. However, it is interesting to continuously evaluate and monitor the volume of care to see whether the downward volume trend in the regional hospital continues and successful substitution will eventually be achieved. To turn the tide, financial incentives and/or payment reform at a regional level need to be considered to increase the financial accountability of the healthcare providers involved.

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08

General Discussion

In order to shift specialised care from the hospital to the primary care setting and to prevent unnecessary referrals to secondary care, the initiative Primary Care Plus (PC+) was implemented in pioneer site 'Blue Care' located in the Maastricht-Heuvelland region in the southern Netherlands. General practitioners (GPs) in the region were able to refer patients with low-complex and non-acute health complaints to PC+ for a maximum of two consultations with a medical specialist from a range of specialties (dermatology, orthopaedics, internal medicine, gastroenterology, neurology, ophthalmology, rheumatology, otorhinolaryngology, paediatrics, gynaecology, urology and a back pain consultation facility). Following a PC+ consultation, patients were referred back to their GP with a diagnosis and/or treatment advice or it was advised to refer the patient to secondary care for further diagnosis and/or treatment. During the whole PC+ process, GPs remained responsible for the patient.

In the same period, the care pathway 'Better exercise in osteoarthritis' was implemented in pioneer site 'Differently Better' located in the Western Mining District of Limburg, which is also located in the South of the Netherlands. Aimed at changing the referral behaviour and diagnostic requesting of GPs, the care pathway stimulated a stepped care approach based on guidelines to optimise the use of existing conservative treatment options and promoted the efficient use of diagnostic imaging for patients with/or suspicion of knee or hip osteoarthritis.

Despite the different approach of these two initiatives, they both aim to prevent and avoid unnecessary care and referrals to secondary care. Therefore, for both interventions, the referral behaviour from primary to secondary care was investigated. Due to the more extensive data available for the PC+ intervention, we were also able to look at the influence of patient and consultation characteristics on this referral decision. Additionally, we studied the effect of PC+ on the outcomes of the Triple Aim. Related to the costs of care, a side-by-side application of the Cost-Utility Analysis (CUA) and Multi-Criteria Decision Analysis (MCDA) was conducted to investigate the applicability and suitability of both methods related to the economic evaluation of new models of care by using PC+ as a case study. Finally, regional healthcare volumes were examined to see how PC+ affects the population level.

In this chapter, the main findings of these studies are presented, followed by a discussion on theoretical and methodological considerations and recommendations for future policy, practice and research.

Main findings

Part I of this dissertation presented several studies conducted to examine the influence of the implementation of PC+ and the care pathway 'Better exercise in osteoarthritis' on the referral behaviour from primary to secondary care and the request of diagnostic imaging. Furthermore, the influence of patient and consultation characteristics on this referral decision was examined. Regarding the influence of PC+ on the referral behaviour from primary to secondary care, it was found that 80.2% of the patients with dermatological complaints were referred back to their GP following PC+ (**Chapter 2**). Therefore, dermatology care seems to be suitable for PC+ and has the ability to reduce hospital based outpatient care referrals. Furthermore, results showed that both the treating specialist and the patient diagnosis independently influenced the referral decision following dermatology care in PC+. Regarding the patient diagnosis, an indication of suitable diagnoses for PC+ was given. For example, due to the large number of patients referred to hospital based outpatient care following PC+, malignant dermatoses are less suitable for PC+. On the other hand, diagnoses like hair and nail disorders, which have low referral rates to outpatient hospital care following PC+, seem particularly suitable for PC+. However, these indications should be handled with care as patients' complaints, symptoms and diseases could ultimately lead to several diagnoses, which are not always known to the GP in advance.

Moreover, these results were supported by the examination of the referral decision following orthopaedic care in PC+ (**Chapter 3**). This study also showed that both treating specialist and the patient diagnosis significantly influenced the referral decision. In addition, to study the effect of the possibility to request diagnostic tests (such as ultrasounds and MRIs) in PC+, referral decisions and consultation- and patient-related factors were compared between two periods: P1 (when it was not possible to request additional diagnostic tests in PC+) and P2 (when the possibility to request additional diagnostic tests was introduced in PC+). Results showed that the possibility to request additional diagnostic tests for orthopaedic surgeons working in PC+ decreased the percentage of referrals to hospital based outpatient care following PC+ (from 39.7% during P1 to 23.7% during P2).

To investigate the effect of the care pathway 'Better exercise in osteoarthritis' on the diagnostic and referral behaviour of GPs, the intervention region was compared with a control region (**Chapter 4**). Results showed that the implementation of the

care pathway using educational meetings, distributing guidelines and incorporating reminders in the GPs' referral application led to a significant decrease in knee-related diagnostics. However, no significant effects were found in hip-related diagnostics, initial orthopaedic consultations and on the probability of undergoing arthroplasty. These mixed effects show that it is important to further investigate the specific role of the different interventions in their effectiveness in improving the diagnostic and referral behaviour of GPs. Furthermore, this study emphasises that diagnostic imaging in primary care is not always necessary to diagnose (in this case) osteoarthritis, which is in contrast to the results of our study related to orthopaedic care in PC+ where medical specialists indicated that diagnostic imaging was a requirement to properly diagnose patients. Therefore, to ensure that overuse of diagnostic tests is not encouraged, it is important to remain critical towards the availability in PC+.

Part II of this dissertation consisted of several studies examining the effect of PC+ on the outcomes of the Triple Aim, on which economic evaluation approach can best be used to decide whether to invest in PC+ and on the effect of PC+ on regional healthcare volumes. Regarding the first two aims of the Triple Aim (population health and patient experiences), patients who visited PC+ (intervention group) and patients who visited hospital based outpatient care (control group) were compared (**Chapter 5**). Results showed that health and quality of care were maintained with PC+. These findings are in line with previous research of Quanjel et al. [1] and confirm that patient's health status, as well as quality of care, are maintained in PC+ since care is provided by the same specialist as in the hospital but closer to patients' homes.

Additionally, PC+ was used as a case study in a side-by-side application of the CUA and the MCDA to compare their applicability and suitability to support local decision makers about the broader value for money of new model of care (**Chapter 6**). To calculate the cost-effectiveness of PC+ using both methods, the health-related quality of life and perceived quality of care outcomes were combined with the costs of care on the individual patient level. Although both methods indicated PC+ as the dominant alternative compared to hospital based outpatient care (care-as-usual), this study concluded that MCDA complements CUA as it provides additional information on a wider range of outcomes and facilitates an auditable and transparent decision process at the local level. However, it was also concluded that more effort is needed to increase the usability of MCDA among local decision makers, as this method comes with several practical challenges.

Despite the positive effect of PC+ at the individual patient level concerning health-related quality of life, perceived quality of care and costs of care as described in Chapter 5 and Chapter 6, it was not clear whether and how PC+ affects regional healthcare volumes at the population level. Therefore, retrospective health insurance reimbursement claims data from the dominant health insurance company in the region were used (**Chapter 7**). With this data, regional care volumes from 2015 to 2018 were compared to the national trend. An increase in the total volume of low-complex specialised care over time in the intervention region was found, by which PC+ did not fully succeed as a substitute for hospital services. Therefore, in addition to measuring interventions' effects on the individual patient level, it was concluded that the effects on the total healthcare utilisation at the population level are necessary to be measured as well.

THEORETICAL REFLECTION

This section reflects on the results of this dissertation. First, a reflection on substitution as a healthcare policy measure and the substitution initiatives included in this dissertation is provided. Second, the effect of substitution initiatives on the healthcare volume at the population level is addressed. Third, a reflection on the behavioural changes required to achieve successful substitution is provided. Finally, the need to move from the Triple Aim to the Quadruple Aim is discussed.

Substitution

In order to be sustainable and to adapt to the increasing demand for healthcare, health systems must provide services that maintain high quality of care and offer better value for money [2]. Using new models of care, healthcare services can be delivered in a different and potentially more cost-effective way through lower cost-providers, locations and formats of delivery [3]. Countries with well-developed primary care systems often opt for strategies aimed at shifting the balance of care from the secondary to primary care setting [4]. By reducing (unnecessary) referrals to and the use of secondary care while reinforcing the gatekeeper role of the GP, substitution of care is a strategy to achieve this balance shift.

The concept of substitution has been used as a policy instrument for healthcare reform for decades, as it was already mentioned by the Netherlands in the Dekker Report [5] in 1987. This report emerged from the White Paper on Primary Care (in Dutch: Nota Eerstelijnszorg) in 1983, which promoted the efficiency of the healthcare

system through a more coherent provision of primary care. Nowadays, the central role of primary care is still emphasised [6], with substitution from secondary care to primary care being an important policy measure [7]. However, substitution does not only refer to the replacement of hospital care by primary care but occurs between and within (care) sectors (i.e. vertical and horizontal substitution) [8]. With substitution, new opportunities changing the actors, the methods, the timing, the location or even the reasons for care are implemented to ensure more effective use of resources.

Substitution mechanisms are at play in different kind of healthcare interventions [9]. First, interventions focus on the care provider delivering the care (e.g. GPs with special interest, nurse practitioners). Second, interventions focus on the setting or location where care is delivered (e.g. shifted outpatient clinics, specialist attachment to primary care teams, telemedicine). Third, interventions focus on the joint management of patients by primary and secondary care professionals (e.g. shared care). Finally, interventions focus on behaviour change of the professional (e.g. guidelines, audit and feedback, financial incentives) or patient (e.g. decision aids). In general, with these initiatives, policymakers aim to facilitate the appropriate delivery of healthcare services at the primary-secondary care interface to overcome fragmentation between providers [9, 10]. In this dissertation, the PC+ intervention, focusing on shifting the venue of specialist care from hospital based outpatient care to the primary care setting without changing the people who deliver the service, is central. In addition, the other intervention discussed in this thesis, the care pathway 'Better exercise in osteoarthritis', can be classified as an intervention that is primarily intended to change the referral behaviour of healthcare professionals (i.e. general practitioners) within the same venue (i.e. primary care centre).

In the feasibility study of van Hoof et al. [11], focusing on specialists performing consultations in GP practices, results showed inefficient and limited use of consultation hours. Besides, close working relationships between GPs and medical specialists in this setting may lead to a relatively low threshold for GPs to refer patients to a medical specialist. Therefore, different from the attachment of specialists to a primary care team, specialists in PC+ work in a neutral primary care setting. Previous research on initiatives where the location of specialist care is shifted to a neutral primary care setting, known as shifted outpatient clinics, shows that these are often popular among patients because of reduced travel time as care is located closer to patients' homes [12, 13] and shorter waiting list time as specialist capacity is expanded [4, 12-16]. Furthermore, positive findings are found

regarding the ability to provide high-quality care for different conditions [4, 12, 14]. These results are in line with the findings presented in this dissertation (Chapter 5). Regarding the costs of care, lower costs per patient were found when specialist services are provided in the primary care setting [4, 12-15, 17] but they come with higher staffing and marginal costs compared to hospital based outpatient care [4, 13, 14, 16]. Furthermore, the cost-effectiveness of this kind of interventions is highly depending on the local arrangements made for reimbursement and the tariffs for the delivery of the service. Due to the diversity of these local arrangements, general guidelines to optimise the configuration of services is difficult. Overall, the number of robust economic evaluations of these types of initiatives is limited, especially those focusing on the whole system [9]. Therefore, this dissertation is not only focusing on individual outcomes related to the Triple Aim but also on the influence of PC+ on the population level.

The care pathway initiative 'Better exercise in osteoarthritis' tried to bring about professional behavioural change through educational meetings, dissemination of guidelines and reminders. Other ways to achieve change are audit and feedback, in-house review of referrals (i.e. second opinion), financial incentives and advice requests. According to the review of Akbari et al. [18] and Giguère et al. [19], passive dissemination of guidelines and other educational materials are ineffective to improve referral behaviour but may have a greater impact when combined or linked with other interventions. For example, implementing referral guidelines supplemented by structured referral sheets or local educational interventions from secondary care specialists are effective in reducing the referral rates without jeopardising the quality of care [9]. Other interventions, such as financial incentives, also proved to lower referral rates but it was unclear how this affected referral appropriateness.

New models of care aimed at substitution continue to develop, for example, by increasingly using nurse practitioners, specialised nurses and physician assistants to provide specialised services in primary care. The introduction of nurse practitioners to chronic care such as diabetes is such an example [20]. In addition, the pandemic of the coronavirus disease-19 (COVID-19) is rapidly introducing the provision of delivering care at a distance using telehealth and e-health [21, 22]. Many of these interventions seem promising but further research is required.

Healthcare volume

An assumption that is often implicit with the introduction of new care models is that they will replace (parts of) existing services [23]. This may indeed be the case if there are so-called 'communicating vessels', in which a higher volume of care in new services is associated with less care utilisation in the existing services (and vice versa) [24]. However, mechanisms exist that new services become complements instead of substitutes, and thereby may add to the overall volume and costs of care [25]. Regarding PC+, a volume increase of low-complex care was indeed observed at the regional population level (Chapter 7).

As stated by Hughes et al. [26], the expectations that integrated care initiatives will improve outcomes and reduce healthcare utilisation at the same time are often disappointing. Since patients and healthcare professionals may have different perceptions of appropriate levels and sources of care, these initiatives can have unexpected consequences, like attracting additional service users [27]. Different mechanisms exist to explain this increase in demand. An often mentioned mechanism in literature is the occurrence of supply-induced demand for healthcare services [28]. In this case, the induced demand is initiated by the supplier (i.e. healthcare provider) and leads to an increase in consultations and/or referrals [28, 29]. Supply-induced demand can be incentivised by substantial information asymmetries between healthcare providers and patients and by the system for financing, organising and paying for medical services. For example, when medical specialists are paid on a fee-for-service basis, they have a financial incentive to over-service patients, compared to specialists paid on a salary basis. Induced demand can also be of interest from the patient perspective, for example, when there is little incentive for patients to restrain demand (moral hazard). Moral hazard refers to the decrease in healthcare consumption when financial consequences are involved [30, 31]. Research showed that the introduction of deductible–based health insurance plans are related to a decrease in primary care and hospital based outpatient care visits [32-34]. Vice versa, supplemental insurance plans appear to be related to an increase in healthcare utilisation [35].

The extent to which there will be an increase in healthcare utilisation is difficult to predict, as a large variation in the use of care across regions and providers is observable [36]. This variation is not only explained by illness, patient risk factors or preferences but can also be explained by the type of healthcare service delivered. For example, supply-sensitive care refers to services where the (local) supply of a

specific resource has a major influence on healthcare utilisation rates. In this case, a higher number of specialist per capita may lead to an increase in healthcare volume. Another category is preference-sensitive care, referring to treatment decisions where different choices bring different benefits and risks, and where the patient's attitude to the outcomes may vary. An example of preference-sensitive care is opting for joint replacement surgery (arthroplasty) in patients with osteoarthritis.

Therefore, to know the impact of a new model of care on the total healthcare utilisation and related costs, evaluations need to take into account the overall healthcare system. When only focusing on the effect of a new model of care on the outcomes at the individual patient level, a limited picture might emerge. Furthermore, successful implementation of substitution initiatives depends on the degree of behavioural engagement of the involved stakeholders (e.g. patients and healthcare professionals) [37].

Behavioural change

According to the results presented in Chapters 2 and 3 of this dissertation, both the treating specialist and the patient diagnosis independently influence the referral decision following dermatology and orthopaedic care in PC+. These results confirm that substitution is more than shifting services out of the hospital and changing the location of healthcare provision [38]. In fact, there is little evidence that just relocation of specialist services to the primary care (or community) settings will reduce dependence on secondary care. New paradigms of care delivery, which are needed when implementing new models of care, require healthcare professionals and patients to do things differently, which involves complex behavioural change [39-41]. This complexity is reflected in the wide variety of behaviours in the different organisations and within and between the different stakeholders involved [42].

A difficulty in healthcare systems is that physician employment does not automatically translate into engaged physicians who are aligned with the aims and activities of their affiliated organisation [43]. Therefore, people must understand the point of the change and agree with it before they actually cooperate in the change in a sustainable way. Phipps-Taylor and Shortell [44] identified a broad range of motivators from research in psychology, organisational behaviour, and industrial psychology that stimulate change in the behaviour of physicians. In their aggregated framework, six domains representing a wide range of motivators are described. First, the domain of social purpose, referring to the joy and satisfaction of doing the

right thing to help patients and colleagues, appears to be particularly prominent in motivation within healthcare settings. In addition, mastery was also considered an important motivator for physicians to engage in change initiatives. This domain refers to the intrinsic joy of learning and using knowledge, teaching others, and monitoring personal improvement. Furthermore, maintaining autonomy and creating a feeling of belonging to an organisation and contributing to shared goals (i.e. relatedness), are two other non-financial motivators. Potential de-motivators or "hygiene" factors are not directly motivating behaviour but can have a positive (or negative) influence on the aggregated effect of the other motivators, for example by reducing workplace stress, employee turnover, and burnout. Finally, direct or associated financial motivators, such as rewards for performing tasks, behaviours, or achieving performance targets, were mentioned as relevant factors to stimulate change. These motivators can be applied to both the GPs and the medical specialists involved in the substitution initiatives described in this dissertation.

Moreover, change is usually achieved through collective action, rather than being a result of individual behavioural processes [42]. Therefore, a 'systems view' on change by using a multilevel change framework is preferred to clarify the structure and dynamics of the total healthcare system. For example, in the framework of Ferlie and Shortell [45] four levels of change are suggested: the individual, the group or team, the overall organisation, and the larger system or environment in which organisations are located. According to this framework, all four levels of change need to be considered to maximise the probability of successful change. First, at the individual level, there are differences in terms of the attitude towards innovation, which is also described by Rogers [46]. However, because healthcare services are delivered in groups or teams within an organisational structure, focusing on changing individuals alone is less effective. Therefore, focusing on groups is a potentially powerful lever for change [47]. As groups of individuals are part of an organisation, they are depending on the complementary resources provided by the organisation to support work and development [48]. An important role of the organisation is therefore to provide an overall climate and culture for change. Finally, change at the organisational level may require reinforcement by macro-level changes in the wider political, economic (or market) environment. Using regulatory, financial, and payment regimes and entities, the structure and performance of healthcare organisations and all other levels of the system can be influenced, directly or indirectly.

On the same note, the importance of contextual factors on different levels influencing change is outlined in the review of Fulop and Robert [49]. According to this approach, a distinction can be made between contextual factors on the micro, meso and macro level, which partly overlap with the four levels of Ferlie and Shortell [45]. At the micro level, contextual factors at the level of one or more professional(s) are of interest, including training, (team) experience and culture. At the meso level, contextual factors within the organisation, like leadership, culture, experience and performance are relevant. Finally, at the macro level, contextual factors include both modifiable and unmodifiable factors. Modifiable factors at the macro level are for example the financial incentives and regulatory mechanism of the healthcare system, levels of competition, and technology. Examples of unmodifiable factors are demography of the population and location in an urban or rural area.

When using a whole system approach, patients should also be included in the process of change. Besides selecting patients most suitable for PC+ using patient profiles, patients also need to be involved actively as they are an important stakeholder in healthcare. When focusing more on patient-centeredness in the delivery of healthcare, the perspective of providers need to be changed and patients and their families should be considered as partners, integrating their values and wishes into the process of care [48]. However, patients and their families are diverse in the level of responsibility and their desire and ability to engage. Therefore, it is important to consider how to tailor efforts to address specific needs and concerns, and best facilitate engagement [39].

Overall, the components that determine the success of substitution initiatives like PC+ and the care pathway are multifactorial and are characterised by a complex interplay [50]. Therefore, it is likely that new models of care are implemented in many different ways. However, keys to successful change and to embed over time are participation and support across all stakeholders and long-term commitment to change [51, 52].

From Triple Aim to Quadruple Aim

The initiatives implemented in the Dutch pioneer sites were focusing on accomplishing the Triple Aim [53]. Therefore, like in many other initiatives, the outcome measures in this dissertation were based on these three aims (i.e. improving population health, improving patient experiences and reducing costs of care). However, as described above, there is a growing realisation among healthcare

organisations that physician involvement is crucial to reform and improve [54, 55]. Moreover, parallel to the effort devoted during the past decades to achieve the Triple Aim, the work environment has worsened, for example by increasing administration requirements, leading to an increasing rate of burnout among healthcare professionals [56, 57]. Because poor morale and burnout may undermine the achievement of the Triple Aim, improving the work-life for healthcare professionals has been added as a fourth aim, resulting in the Quadruple Aim [58]. By adding this fourth dimension, positive engagement of the healthcare workforce is emphasised [59]. An engaged, healthy workforce that delivers high-quality patient care ultimately facilitates improved patient experiences, population health and lower costs [58]. With the Quadruple Aim, conditions are created for the healthcare workforce to find joy and meaning in their work and thereby improving the experience of providing care.

Although not reflected in the previous chapters of this dissertation, attention was paid to this fourth aim during the evaluation of the substitution initiatives. In a Dutch report including the results of the monitoring and evaluation of the three pioneer sites in Limburg, a concise study focusing on the experience of healthcare professionals with PC+ was published [60]. Results of this quantitative questionnaire study showed that PC+ positively affects the job satisfaction of medical specialists and indicated that they appreciate the atmosphere in PC+. However, this research also showed that GPs feel less involved in PC+ and that contact and collaboration between GPs and medical specialists were not optimal. In addition, in an exploration of the possibilities for further development of PC+, healthcare professionals as well as other stakeholders, were interviewed [61]. This qualitative study asked about the opinion of healthcare professionals about their vision of PC+, their experiences with PC+ and their opinion about the possibilities for renewal and expansion of PC+. Continuous involvement of healthcare professionals in evaluating and monitoring the PC+ initiative is important because they have a key position concerning the degree of success.

METHODOLOGICAL REFLECTION

This section contains a reflection on the different methodological strengths and limitations of the studies presented in this dissertation. First, the study design is discussed. Second, the different data sources and analysis methods used are addressed. Third, the different outcomes and levels of outcomes are reflected upon. Finally, attention is paid to the generalisability of this dissertation.

Study design

This dissertation consists of several studies with an observational design. Despite that in most circumstances experimental designs are preferred to observational designs, they are not always practicable and feasible, for example, because large scale implementation of an intervention is already underway, withholding an intervention for the control group is considered unethical, or because insufficient external validity threatens the generalisability of findings to clinical relevance and the general population [62-65]. In the case of the substitution initiatives described in this dissertation, the complex systems in which these interventions are implemented make an experimental design less practicable. As mentioned by Shiell et al. [66], complex systems interaction occurs between components of the intervention as well as between the intervention and the context in which it is implemented. Furthermore, since well-designed observational studies are increasingly able to provide valid results, they are becoming more popular [67].

Despite the increasing use of observational study designs, the presence of bias is an often discussed issue, like selection bias and confounding [68, 69]. Selection bias occurs when patient participation in an intervention is related to exposures or outcomes of interest. For example, in PC+ the allocation of patients to the intervention group (PC+) or control group (hospital based outpatient care) was not at random but based on the decision of the GP in interaction with the patient. Based on clinical expertise and shared decision-making, GPs decided whether to refer a patient to PC+. When GPs that do refer patients to PC+ differ from and/or serve different patients than GPs who do not, or to a lesser extent, refer patients to PC+, selection bias across the intervention and control group may occur [70]. In addition, selection bias also occurs when the intervention and control group themselves change in composition across time. Furthermore, confounding refers to the possibility that measured and unmeasured factors have incomparable frequencies in the intervention and control group due to the non-random allocation of patients [71]. To minimise selection bias and to control for confounding, propensity score matching was used in this dissertation (Chapter 5 and 6). The use of propensity scores was first introduced by Rosenbaum and Rubin (1983) [72] aiming to make the intervention and control group as similar as possible concerning the observed baseline characteristics. Nowadays, propensity score matching is a popular and powerful technology that allows for causal inference in observational study designs without randomisation of patients to an intervention or control group [72]. However, researchers must be aware of the limitations and carefully interpret and present the findings. One of

these limitations is that propensity score matching can be used to correct overt bias, which results from measured variables, but not for hidden bias, which results from unobserved and unmeasured variables [73].

In addition to observational study designs, other non-experimental alternatives can be considered when evaluating new models of care like PC+ and care pathways. Different quasi- and non-experimental study designs are suitable, like natural experiments in which experimental thinking is applied to non-experimental situations [74]. Another approach for the evaluation of new models of care is the realist evaluation approach of Pawson and Tilley (1997) [75]. According to this approach, the intervention outcomes are generated by mechanisms which are triggered in a particular context through an actor or actors (i.e. stakeholder(s)) involved [76]. Therefore, the evaluation of interventions should not only focus on the impact of the intervention but should also address 'what works, how, for whom, and in what circumstances'. Nevertheless, when an experimental design is preferred, random allocation does not have to be limited to individuals, it can also be applied at a group level by randomising one or more clusters, for example, GP practices [77, 78].

Finally, the studies described in this dissertation are all quantitative. Although a qualitative study is missing, qualitative research has been conducted during the evaluation of the substitution initiatives. For example, the patient representative foundation 'Burgerkracht Limburg' interviewed patients to monitor the quality of care in PC+. In addition, healthcare professionals and patients involved in the care pathway 'Better exercise in osteoarthritis' were also interviewed about their opinion about osteoarthritis-related care. Furthermore, after the evaluation of PC+ interviews were held with medical specialists working in PC+, GPs and the initiators (the primary care organisation 'Care in Development', the Maastricht University Medical Centre+, the health insurance company VGZ, and the patient representative foundation 'Burgerkracht Limburg') in order to identify needs for improvement [61]. This qualitative follow-up has been used to reflect upon the quantitative findings of the evaluation [60]. Besides interviews, there have always been feedback moments between the researchers and the stakeholders of the initiatives. For example, the expert meetings used in the different studies to verify study findings and to contribute to a better interpretation of the results. These qualitative additions have yielded a great deal of knowledge; however, a stronger embedding of qualitative methods in the various studies would be a good addition.

Data sources and methodologies

To study the effect of the two substitution initiatives (PC+ and the care pathway 'Better exercise in osteoarthritis') on diagnostic requests and number of referrals to secondary care and the outcomes of the Triple Aim and regional healthcare volumes, different data sources and methodologies were used. The various data sources used in this dissertation are retrospective monitoring data from the PC+ centres (Chapter 2, 3 and 6), health insurance reimbursement claims data (Chapter 4 and 7), patient questionnaires (Chapter 5 and 6), and PC+ and hospital administrative data (Chapter 6). By using these different sources, the research questions have been approached from various perspectives.

Monitoring data, claims data and administrative data are representing the actual care that patients receive. These data sources consist of data collected without the presence and influence of researchers and study protocols. With these sources, accurate data for a large sample can be obtained at a low cost [79]. When conducting the CUA and MCDA (Chapter 6), patient quality of life and experience of care needed to be linked with the costs of care and therefore the patients' questionnaire data were linked to the PC+ and hospital administrative datasets. By drawing the costs of care from the PC+ and hospital datasets, the burden on respondents, survey dropouts and item non-response rates were reduced [80]. Data linkage preferably takes place based on a unique identifier (e.g. a citizen service number, tax or social insurance number). However, such an identifier was not included in the data sources used in the studies described in this dissertation. Therefore, data linkage was based on variables such as gender and date of birth. However, because of inconsistencies, inaccuracies and duplications in the data sources, perfect data linkage was not possible resulting in the loss of respondents and therefore resulted in different patient populations in the different studies included in this dissertation (Chapter 5 and 6). Furthermore, with the introduction of the General Data Protection Regulation (in Dutch: Algemene verordening gegevensbescherming) in 2018, data linkage between patient questionnaire data and health insurance reimbursement claims data was no longer possible. This resulted in limited insight into the costs of care because only PC+ and the hospital care related costs could be included. However, it is assumed that these categories are likely to account for the largest share of the total costs.

Besides different data sources, various methodologies are used to analyse the data and to answer the different research questions in this dissertation. For the research on referral decisions from patients who received dermatology care (Chapter 2) and

orthopaedic care (Chapter 3) in PC+, (stepwise) logistic regression modelling was used to describe variations in this referral decision. By using logistic regression analysis, the relationship between different variables (or predictors) on the referral decision following PC+ can be evaluated. However, choosing the right predictors is key to a successful model. Herewith, it is important to include only relevant variables (i.e. scientific plausible), as otherwise associations can be diluted, large standard errors with wide and imprecise confidence intervals may arise, or false associations may be identified [81]. Results of these studies showed that both specialists and patients significantly influenced the referral decision following PC+. This information can be used to develop appropriate profiles to indicate suitable specialists and patients for PC+. Furthermore, to evaluate the effect of the care pathway on the diagnostic and referral behaviour of GPs, binary logistic regression analysis was applied (Chapter 4). The mixed results of this study indicated the need to identify the specific role of the different intervention parts in their effectiveness.

Additionally, to compare patients referred to PC+ and patients referred to hospital based outpatient care on the Triple Aim outcomes, propensity score matching to control for potential (overt) selection bias was used (Chapter 5 and 6). As described earlier, this technique allows making the intervention and control groups as comparable as possible concerning the observed baseline characteristics. From this, it has been concluded that population health and quality of care are maintained in PC+ (Chapter 5). Besides this, the matched groups were used to evaluate the cost-effectiveness of PC+ using the CUA and MCDA (Chapter 6). Both methods indicated PC+ as less costly and more effective compared to care-as-usual. Furthermore, a side-by-side application of these two methods was applied to compare their applicability and suitability. This comparison showed that MCDA may complement CUA to provide additional information on a wider range of outcomes and facilitate an auditable and transparent decision process at the local level. However, it should be noted that criteria weights of the SELFIE study were used [82], which included people with more complex healthcare needs. Although this was not ideal, the impact on the total performance scores in the MCDA are expected to be limited because the SELFIE study used general outcome concepts (grouped by the Triple Aim) to define the criteria. Furthermore, the SELFIE criteria weights were derived from approximately 180 Dutch stakeholders including patients, partners of patients, providers, payers, and policy makers from across the Netherlands. Therefore, it was possible to reuse the SELFIE framework for the case study of PC+.

Finally, to study the effect of PC+ on the healthcare volumes at the population level, regional trends were described and compared to the national trend (Chapter 7). Results showed an increase in the total volume of low-complex specialised care in the intervention region over time, which was not visible at the national level. Therefore, it was concluded that despite the positive outcomes of PC+ at the individual patient level, desired results related to the volume of low-complex specialised care at the population level are limited. Although these trend analyses do not validate the actual influence of PC+ on the healthcare utilisation (i.e. causal inference), it does indicate that it is important to evaluate outcomes at the population level and to continuously evaluate and monitor the intervention on both the individual and population levels.

By using different data sources and different methods, this dissertation provides more insight into the influence of PC+ and the care pathway on the referral behaviour from primary to secondary care and the request of diagnostic imaging. Furthermore, it has also led to more clarity about the effect of PC+ on the whole system, including the outcomes at the patient level (Triple Aim) and the population level (volume of care).

Outcomes

To evaluate whether the intended goals of the substitution initiatives are being achieved and which factors determine success, outcomes concerning both process and effect evaluation are valuable. By evaluating the referral decision following dermatological and orthopaedic care in PC+ (Chapter 2 and 3), insights have been gained into which facilitating and hindering factors are relevant for PC+. Furthermore, the evaluation of the care pathway (Chapter 4) focused on outcomes related to the effect on the diagnostic and referral behaviour of GPs and the number of hip and knee arthroplasties but also gave insights into the practical application of the pathway. In addition, the final three studies (Chapter 5, 6 and 7) focused on the effect of PC+ on the Triple Aim outcomes and the volume of care.

To measure the influence of PC+ and the care pathway on the referral behaviour and request of diagnostic imaging, outcomes were selected based on the availability of variables in the different data sets. As a consequence, the number of factors included to predict the referral decision following PC+ was limited. To optimise these predictions, additional information, especially from the GPs' perspective, would be necessary. With more accurate predictions, more information to optimise the PC+ process could be provided. Regarding the effect of PC+ on the Triple Aim, this dissertation defines all three dimensions, resulting in the conclusion that, compared

to care-as-usual, PC+ is able to deliver the same quality of care with comparable outcomes on health-related quality of life with lower costs-per-patient. When adding the results of the monitoring and evaluation report [60], the outcomes on the fourth aim, improving the work-life for healthcare professionals, can be covered as well. The addition that PC+ positively affects the job satisfaction of medical specialists, provides an even more complete picture. Furthermore, when using the Triple or Quadruple Aim framework to evaluate new models of care, a wide variety of outcome measures is available. This can make it difficult to select the relevant outcome measures. However, often this selection depends on the data availability, resource constraints, and overall objectives [83]. This was also the case in the evaluation of PC+ as the choice of indicators was tuned with the National Institute for Public Health and the Environment (in Dutch: Rijksinstituut voor Volksgezondheid en Milieu, RIVM) to make comparison of the pioneer sites possible. Moreover, in addition to generic questionnaires, specialty and/or condition-specific questionnaires could be used. To do so, it is recommended to use existing valid and reliable questionnaires as much as possible. In addition to questionnaires, it may be considered to measure the health of the population with clinical outcome measures specific to the complaint or condition which are registered in PC+ and/or the hospital.

Besides focusing on the outcomes at the individual patient level based on the Triple Aim, this dissertation also includes outcomes at the population level. Results focusing on the volume of care show that positive findings at the individual patient level do not automatically lead to a positive effect (i.e. substitution of care) at the population level. This implies that although PC+ is cheaper compared to care-as-usual, it does not automatically lead to cost savings for the whole region. Because of the absence of claims data related to primary care, insight into the use of primary care following PC+ is lacking. Nevertheless, the impact of the use of primary care in terms of costs in primary care will be limited because these costs are substantially lower compared to secondary care.

Generalisability

At the national level, the findings of this dissertation are to a great extent generalisable to other regions in the Netherlands. However, it is important to realise that new models of care are built on old foundations [84]. Therefore, the context, both historical and current, must be understood before implementing new models of care. The pioneer sites ‘Blue Care’ and ‘Differently Better’ are characterised by a long history of collaboration between the different healthcare organisations, like the

regional care group, the regional hospital, the patient representative foundation and the dominant healthcare insurer. Close collaboration between these stakeholders is of high importance when implementing substitution initiatives. Furthermore, the complexity of the region also plays an important role in this collaboration. In the pioneer sites 'Blue Care' and 'Differently Better', the populations are delineated by patients registered with GPs belonging to one particular care group and for each of the regions, only one hospital is located. This may lead to less conflict of interests and contributes to a more manageable collaboration. Although the new care models presented in this dissertation can be viewed as blueprints for other regions to learn from, it is necessary to adapt the initiatives and their elements to the regions before they are implemented. Therefore, each region should take time to understand and adapt to the relevant context and for people and organisations to collaborate.

Regarding the generalisability of the findings in this dissertation on the global level, it is important to note that the Dutch healthcare system differs from other countries. Both the PC+ and the care pathway initiative focus on strengthening primary care as it serves as a patient's entry point to the healthcare system in the Netherlands. Spain, Italy and the UK are examples of countries with a similar 'gatekeeper' system in which primary care controls access to most types of secondary care [85]. Among others, in Belgium, France and Denmark patients have direct access to secondary care without a GP referral. Despite these different healthcare systems, there is increasing emphasis on the need to strengthen primary care to better address the needs of the (ageing) population and to reduce the unnecessary use of hospital care (ref). Therefore, substitution initiatives could be implemented in many different healthcare systems.

FUTURE DIRECTIONS

Based on the results presented in this dissertation, recommendations for policy, practice and further research are given.

Policy

PC+ and the care pathway 'Better exercise in osteoarthritis' are both implemented in a so-called pioneer site. These pioneer sites were initiated by the Dutch Ministry of Health, Welfare and Sport in 2013 in a nationwide effort to achieve 'better healthcare at lower cost' [53]. The pioneer sites were characterised by a bottom-up approach in which stakeholders together took responsibility to achieve better and affordable care.

In line with the goal of the pioneer sites, the Ministry of Health, Welfare and Sport and stakeholders of medical care, like the Dutch Association of Hospitals, the Dutch organisation of Health Insurers and The Netherlands Patients Federation signed the 2019-2022 framework agreement in 2018 (in Dutch: hoofdlijnenakkoord) [86]. This agreement is a combination of controlling expenditure on the one hand and guaranteeing accessibility and affordability of healthcare in the long term on the other. Eventually, the aim is to slow down the growth in expenditure on specialised medical care to 0% in 2022. In order to achieve this reduction, incidental money is available for organisations to focus on 'the right care in the right place'. 'The right care in the right place' is a movement to support the sustainability of healthcare and focusses on avoiding expensive care, moving the point of care delivery closer to people's homes and replacing care delivery with other forms such as e-health [87].

Regarding the initiatives described in this dissertation, the next step is to further develop and optimise them in accordance with these new agreements. Regarding the PC+ initiative, we explored the possibilities for further development and optimisation together with the stakeholders involved [61]. Themes discussed during this exploration are for example the vision and position of PC+, referral behaviour of GPs, communication and collaboration between healthcare professionals, and the expansion of healthcare professionals, patient groups and medical specialties in PC+. These items will be discussed further in the practical recommendations of this chapter.

Another important point for policy is the realisation that when shifting care out of the hospital, implementation challenges are significant and even initiatives with great potential might fail [88]. This is often because there is not enough realisation that the intended shift in care cannot be achieved without significantly increasing

capacity and competence in primary and community care and solving some of the prevailing problems in social care. Therefore, a wide range of system, organisational and individual factors that impact feasibility and effectiveness should be taken into account. In line with this whole system approach, shifting to integrated payment mechanisms intended to incentivise high-value care may be necessary to achieve better patient outcomes and lower costs [26]. Common payment models like fee-for-service or case payment (e.g. diagnostic related groups) are promoting volume and taking little account for value [50]. Therefore, to make sure that new models of care improve value, new payment models need to create incentives for behavioural change among healthcare providers and patients [89]. Additionally, new contractual arrangements between health insurers and providers are needed to overcome the 'silo effect' and fragmentation of care [50]. Instead, these arrangements should focus on providing care across organisations for a total population [90]. Such a geographical population health focus is essential to take into account the characteristics of the macro-level context in order to maximise patient accessibility and minimise duplication of services [91]. Furthermore, in order to achieve successful substitution of care, financial agreements between health insurers and healthcare organisations should be transparent. All stakeholders should be aware of the implications of these agreements. In secondary care, substitution may lead to a decrease in both the volume of patients and the volume of care delivered. In addition, in primary care, it must be clear which patients can be referred to PC+, and unnecessary referrals to PC+ should be avoided.

Practice

The results of the studies focusing on the referral decision following PC+ (Chapter 2 and 3) showed that both specialists and patients have a significant influence. Regarding the profile of medical specialists working in PC+, van Hoof et al. [11] described that medical specialists should have a certain degree of seniority, work according to a generalist approach, and have an attitude consistent with the model of substitution. Developing such a profile, describing the qualifications of an eligible medical specialist, is considered as a precondition for PC+. The results of this dissertation build on these findings and agree that the ability to work in a PC+ setting differs among specialists. Because it is expected that medical specialists with a less generalist approach may refer patients to outpatient hospital care more often, or even do not like to work in such a setting, more research is needed to study the most appropriate profile of specialists to work in PC+. Furthermore, it may be an

opportunity to train specialists with an affinity to work in these settings as well as to train physicians educated to become a specialist during internships in these settings.

Regarding the influence of patients on the referral decision following PC+, both studies (Chapter 2 and 3) provide indications of diagnoses that are, to a greater or lesser extent, suitable for PC+. These results provide input for the further development of patient profiles to better support GPs in deciding whether to refer a patient to PC+. In addition, it is recommended to continuously evaluate these profiles by GPs and medical specialists and to make adjustments when necessary. Herewith, it is important to take into account the working methods of GPs and to process the patient profiles in referral forms and/or referral systems as a constant reminder.

Another issue that was relevant in examining the referral decision following PC+ was the influence of diagnostic tests on this decision (Chapter 3). Results showed that the possibility to request additional diagnostic tests for orthopaedic care in PC+ had a significant impact on the referral decision. Despite these positive findings, it was also food for thought whether the availability of diagnostic tests fits within the vision of PC+. In order to prevent substitution initiatives from being nothing more than relocated hospital care, it is important to ascertain whether and which diagnostic tools should be available for each medical specialty to enable the establishment of an accurate diagnosis. In addition, it is recommended to arrange the diagnostic possibilities as efficiently as possible for the patient. For example, by offering the diagnostics in the PC+ centre. Besides this, to enable diagnostic tools in PC+, it can be taken as a starting point that only primary care diagnostics are available. With this, the vision that the GP remains in control and the medical specialist provides treatment advice is honoured.

An important issue related to the care pathway (Chapter 4) was the referral behaviour of GPs. Results showed that not all GPs seem to conform their referral behaviour to the care pathway. This lack of conformity was also reflected in the data from VGZ, showing varying referral behaviour from GPs to PC+ [61] and in the results of the questionnaire study focusing on the experience of healthcare professionals with PC+, showing that less than half of the GPs felt involved in the PC+ initiative [60]. The goal of substitution initiatives is to bridge the gap between primary and secondary care and therefore the involvement of both GPs and medical specialists is important. However, GPs with many referrals to PC+ do not automatically achieve substitution of care as they might experience a (too) low threshold [61]. Therefore, it is important

to remain critical about the accuracy of the referrals in order to avoid unnecessary referrals. A solution proposed by the GPs involved is discussing the necessity of the referrals during 'intervision meetings' within their own GP practice. Another example is organising speed dates between GPs with many and few referrals to discuss their differences which is an intervention of the 'Plus Practices', another initiative in South Limburg [92]. By providing 'mirror information', GPs' awareness of their gatekeeper role is increased and guidance is provided for fulfilling this role.

Another important point is the communication between GPs and specialists in substitution initiatives. Although specialists performing consultations in GP practices resulted in (too) close working relationships between healthcare professionals [11], in the current working method of PC+, contact, cooperation and knowledge sharing between healthcare professionals is lagging. This complies with previous research of Moffatt et al. [93] in which they mention that the attachment of specialists to primary care teams seem to have a stronger educational focus than shifted outpatient clinics. According to Winpenny et al. [9] information about how outpatient clinic initiatives affect the interaction between GPs and medical specialists and if and how they create a learning effect is lacking. Regarding the care pathway, it was also concluded that collaboration and communication between healthcare professionals can be improved. To further stimulate collaboration among healthcare professionals, they must get to know each other on a more personal level [94]. Besides that knowing each other makes cooperation more enjoyable, more candid, and easier, it also leads to a better understanding of each other's working methods and relevant medical competences. Suggestions made by the stakeholders involved in the pioneer sites to facilitate this is to organise so called 'meet-and-greet' events for GPs and medical specialists, and joint training sessions. To further optimise communication and collaboration, shorter lines between GPs and medical specialists should be created so that advice can be requested in an accessible way (for example by e-mail or by telephone). In addition, attention can also be paid to the quality of the referral letters. This concerns communication from the GP to the medical specialist as well as feedback from the medical specialist back to the GP.

Additionally, the deployment of other professionals (e.g. GPs with special interest, nurse practitioners, specialised nurses and physician assistants) is another important point regarding the further development of PC+. Despite diverse experiments with other professionals in PC+ (e.g. GP with a special interest in gynaecology, nurse-led stroke aftercare) its feasibility has not yet been investigated.

The suitability of these professionals must be determined for each specialism and concerning specific patient profiles and/or diagnostic groups. Eventually, the deployment of other professionals could lead to more cost savings, as the hourly rate of these healthcare professionals is lower than that of a medical specialist. However, it is recommended to implement this deployment under the supervision of a medical specialist, since this is considered of additional value by patients [95]. Furthermore, PC+ is focusing on patients with low complex complaints who would otherwise have been referred by the GP to hospital based outpatient care. Herewith, PC+ aims to avoid referrals to secondary care. However, the vision of PC+ could also be expanded and also focus on other patients groups, like following-up patients with particular chronic conditions or multimorbid patients. The evaluation of nurse-led stroke aftercare in PC+ showed positive results regarding the cost-effectiveness of this form of reverse substitution [ref. Daan Verberne]. In addition, a physical consultation with a healthcare professional may not always be necessary. E-health could also be used within some medical specialties, such as teleconsultations. By using e-health, waiting lists can be reduced and costs can be saved [96]. Especially during the current COVID-19 pandemic, the relevance of the provision of care at a distance is high and may therefore be implemented more quickly.

Research

When initiating new models of care, it is important to have a clear plan about the method of monitoring and evaluation and to involve researchers in the process prior to the implementation of initiatives. Besides this, after the implementation, continuous monitoring and evaluation is important because these initiatives are constantly adapted to new insights and/or circumstances. This process remains important even after a pilot phase as the substitution initiatives are still developing and improving, for example, with the deployment of other professionals, the implementation of e-health, and the inclusion of other patients groups. The outcomes on the Triple Aim, or the Quadruple Aim, continue to play an important role in evaluating the success of the initiatives.

Additionally, according to a realist approach, additional research should also focus on the 'programme mechanisms' of the substitution initiatives [97]. These mechanisms play a role in understanding the interaction between what the intervention provides and the reasoning of the actors that influences the outcomes. For example, the hospital administrative data showed differences in the substitution effect between the involved medical specialties [60]. Therefore, more research is

needed to understand what works for which medical specialty and what mechanisms are triggered. In that perspective, the various working and/or referral methods (for instance using triage), of the various medical specialties involved could be an interesting issue for further research. Besides this, more focus on the influence of the GPs involved is recommended because research with VGZ claims data showed that the referral pattern to a medical specialist varies among the GPs [61]. Some elements of a realist impact evaluation were already present in the research into the substitution initiatives, but this can be made more explicit. Embedding qualitative research to gain more insight into the underlying mechanisms of successful application and adaptation of healthcare professionals to the intervention can be a good addition to this. By using qualitative research, more attention can be paid to examples of best practices from the various medical specialties involved. In addition, it can provide more insight into the referral culture and referral behaviour of the different GPs and within the different GP practices.

Furthermore, there is still room for improvement in the field of data collection and linkage. For example, to elaborate on the 'programme mechanisms', all necessary information must be registered to gain insight into the processes and results of care. However, at the moment the health insurance claims data do not show to which medical specialty a patient was referred in PC+. Therefore, it is difficult to get an overview of the total course of care per medical specialty (i.e. care use in primary care, PC+ and secondary care) and only conclusions can be drawn at a general level (for all medical specialties together). Especially when medical specialties start experimenting with, for example, the deployment of other healthcare professionals in PC+, it would be informative to get a clear picture of the effect on all levels of care.

Together, this information should give understanding about 'what works for whom, in what contexts, and how' for the different initiatives. This information can be useful for other regions, since new models of care work differently in different contexts and through different change mechanisms resulting in different outcomes. Therefore, by using realist impact evaluation, more information about mechanisms and contexts useful to policy and practice can be provided. In this way, the results of PC+ and the care pathway can serve as blueprints for other regions to learn from.

Finally, the absence of the desired substitution effect immediately after implementation does not mean that the initiatives do not affect. Substitution initiatives need time to develop, to generate sufficient volume and effect, and to allow

healthcare professionals to make required behavioural adjustments. Therefore, a longitudinal approach is recommended.

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Summary

In response to fragmentation and care coordination problems in healthcare and to transform healthcare delivery, health authorities worldwide are experimenting with new models of care at local level. These so-called pilot sites, pioneer sites, or vanguards act as blueprints and inspiration to the rest of the healthcare system. Also in the Netherlands, improving the financial sustainability of the healthcare system is high on the political agenda. Therefore, regional innovation initiatives are designated as pioneer sites in a nationwide effort to improve the health of the population and quality of care, and to limit the growth of healthcare expenditure (Triple Aim). One of the strategies used in these pioneer sites is to provide more integrated care as well as increasing efficiency of care by shifting specialised medical care from hospital based outpatient care to primary care without changing the people who deliver the service, as an example of substitution of care. Primary Care Plus (PC+) implemented in pioneer site 'Blue Care' located in the Maastricht-Heuvelland region is an example of a substitution initiative focusing on medical specialists providing face-to-face consultations in a primary care setting. In the same period, the care pathway 'Better exercise in osteoarthritis' was implemented in pioneer site 'Differently Better' located in the Western Mining District of Limburg. Aimed at changing the referral behaviour and diagnostic requesting of general practitioners (GPs), the care pathway stimulated a stepped care approach based on guidelines to optimise the use of existing conservative treatment options and to promote the efficient use of diagnostic imaging for patients with/or suspicion of knee or hip osteoarthritis. Despite the different approach of these two initiatives, they both aim to prevent and avoid unnecessary care and referrals to secondary care.

Chapter 1 introduces the topic of this dissertation. It provides background information about relevant healthcare issues like fragmentation of care and rising healthcare costs. Additionally, it elaborates on approaches to overcome these problems, such as integrated care and patient-centred medicine. Furthermore, the development of new models of care as a response to these problems is described, with a focus on the Dutch substitution initiatives PC+ and the care pathway 'Better exercise in osteoarthritis'. This is followed by the overall aims of this study: (1) to examine the effect of both initiatives on the referral behaviour to secondary care and/or for diagnostic imaging and to identify the influence of predictive characteristics on the decision whether to refer to secondary care, and (2) to examine the effect of PC+ on patients' health-related

quality of life, the patients' experienced quality of care, the cost-effectiveness, and the volume of care on a regional level.

Chapter 2 explores the referral decisions following dermatology care in PC+ and the influence of predictive patient and consultation characteristics on this decision. In this retrospective study, clinical data of patients who received dermatology care in PC+ between January 2015 and March 2017 are used. The referral decision following PC+, (i.e., referral back to the GP or referral to outpatient hospital care) was the primary outcome. Stepwise logistic regression modelling was used to describe variations in the referral decisions following PC+, with patient age and gender, number of PC+ consultations, patient diagnosis and treatment specialist as the predicting factors. Results showed that 80.2% of the patients who visited PC+ for dermatology care were referred back to the GP. Therefore, dermatology care seems to be suitable for PC+ and has the ability to reduce hospital based outpatient care referrals. Furthermore, results showed that both the treating specialist and the patient diagnosis independently influenced the referral decision following dermatology care in PC+. Regarding the patient diagnosis, an indication of suitable diagnoses for PC+ was given. For example, due to the large number of patients referred to hospital based outpatient care following PC+, malignant dermatoses are less suitable for PC+. On the other hand, diagnoses like hair and nail disorders, which have low referral rates to outpatient hospital care following PC+, seem particularly suitable for PC+. However, these indications should be handled with care because patients' complaints, symptoms and diseases could ultimately lead to several diagnoses, which are not always known in advance by the GP. Overall, the results of this study can be used to discuss and improve specialist and patient profiles for PC+ to further optimise the effectiveness of the initiative.

Chapter 3 describes the evaluation of the referral decision following orthopaedic care in PC+ and in particular the evaluation of the influence of diagnostic tests on this decision. Therefore, retrospective monitoring data of patients visiting PC+ for orthopaedic care were used. The primary outcome was the referral decision following PC+ (back to the GP or referral to outpatient hospital care). Independent variables were consultation- and patient-related predictors. To describe variations in the referral decision, logistic regression modelling was used. In line with the results of dermatological care in PC+, this study also showed that both treating specialist and the patient diagnosis significantly influenced the referral decision. In addition, to study the effect of the possibility to request diagnostic tests (such as ultrasounds and MRIs) in PC+, referral decisions and consultation- and patient-related factors were compared between two

periods: P1 (when it was not possible to request additional diagnostic tests in PC+) and P2 (when the possibility to request additional diagnostic tests was introduced in PC+). Results showed that the possibility of requesting additional diagnostic tests for orthopaedic surgeons working in PC+ significantly decreased the number of referrals to hospital based outpatient care. Despite the significant impact of the possibility of requesting additional diagnostic tests in PC+, it is important to discuss the extent to which the availability of diagnostic tests fits within the vision of PC+. In addition, selecting appropriate profiles for specialists and patients for PC+ are necessary to further optimise the effectiveness and cost of care.

Chapter 4 presents the results of the study on the stepped-care approach, in the shape of the care pathway 'Better exercise in osteoarthritis', which was implemented in the pioneer site 'Differently Better' to reduce the number of diagnostic imaging requested by GPs and referrals of GPs to orthopaedic care. In 2015, the pathway is implemented with the use of educational meetings, distributing guidelines and incorporating reminders in the GPs' referral application. To evaluate the effect of the pathway on the diagnostic and referral behaviour of GPs, hip and knee related health insurance claims are used together with claims of other joints and of a control region for comparison. The average number of claims and the percentage change in the post-implementation period are described. Binary logistic regression analysis is used to examine the interaction between region (intervention and control) and period (pre- and post-implementation). Using random sampling of patient records, information about the practical application of the pathway and the number of hip or knee arthroplasties is added. Results showed that the implementation of the care pathway led to a significant decrease in knee-related diagnostics. However, no significant effects were found in hip-related diagnostics, initial orthopaedic consultations and on the probability of undergoing arthroplasty. Therefore, the referral behaviour of GPs to orthopaedic care needs attention for future interventions and research, since an increase (instead of a desired decrease) in the number of initial orthopaedic consultations was found. Focusing on the entire width of care for hip and knee osteoarthritis could be helpful. Furthermore, this study emphasised that diagnostic imaging in primary care is not always necessary to diagnose (in this case) osteoarthritis, which is in contrast to the results of our study related to orthopaedic care in PC+ were medical specialists indicated that diagnostic imaging was a requirement to properly diagnose patients.

Chapter 5 shows whether population health and experience of care in PC+ could be maintained. To do this, health-related quality of life and experienced quality of care

from a patient perspective were compared between patients referred to PC+ and to hospital-based outpatient care (care-as-usual). In this cohort study, patients from the Maastricht–Heuvelland region visiting PC+ or hospital-based outpatient care between December 2014 and April 2018 were included. With patient questionnaires (To, T1 and T2), the health-related quality of life and experience of care were measured. One-to-two nearest neighbour calliper propensity score matching was used to control for potential selection bias. Outcomes were compared using marginal linear models and Pearson chi-square tests. Results showed that health and quality of care were maintained with PC+. Only significant differences in travel time between the intervention and control group were found. Therefore, this study concluded that future research should focus more on cost-related outcomes.

Chapter 6 presents a side-by-side application of the Cost-Utility Analysis (CUA) and Multi-Criteria Decision Analysis (MCDA) to investigate the applicability and suitability of both methods to support local decision makers about the broader value for money of new model of care. By using PC+ as a case study, it is investigated whether the adoption of MCDA instead of traditional CUA alters the decision of investing in PC+. Data of patients referred to PC+ or care-as-usual were retrieved by questionnaires and administrative databases with a three-month follow-up. Propensity score matching together with generalised linear regression models was used to reduce confounding. Univariate and probabilistic sensitivity analyses were performed to explore uncertainty in the results. Although both methods indicated PC+ as dominant alternative, complementary differences were observed. MCDA provided additional evidence that (1) PC+ improved access to care, (2) improvement in health-related quality of life was driven by the psychological well-being component, and (3) the budget required for PC+ to be affordable in addition to preferable was estimated at €521.42 per patient. Additionally, MCDA was less sensitive to the utility measures used. Therefore, MCDA may facilitate an auditable and transparent evaluation of new models of care by providing additional information on a wider range of outcomes and incorporating affordability. However, more effort is needed to increase the usability of MCDA among local decision makers.

Chapter 7 describes whether and how PC+ affects regional healthcare volumes at the population level. Previous research describes possible mechanisms by which PC+ could be a substitution for or an addition to other types of health services. Therefore, this study used retrospective health insurance reimbursement claims data from the dominant health insurance company in the region to determine regional

care volumes from 2015 to 2018 and compare them to the national trend. The results show an increase in the total volume of low-complex specialised care over time in the intervention region, by which PC+ did not fully succeed as a substitute for hospital services. Therefore, in addition to measuring interventions' effects on the individual patient level, it was concluded that the effects on the total healthcare utilisation at the population level are necessary to be measured as well. Furthermore, time is needed for this kind of intervention to be effective based on the results of continuous monitoring and evaluation and hence adaptation. Thus, a longitudinal dynamic evaluation approach to inform policy makers is needed.

Finally a summary of the main findings of this dissertation is given, followed by a reflection on the theoretical and methodological considerations. Furthermore, implications for policy, practice and research are discussed.

Samenvatting

Als reactie op fragmentatie en coördinatieproblemen in de gezondheidszorg en om de zorg te transformeren, experimenteren gezondheidsautoriteiten wereldwijd met nieuwe zorgmodellen op lokaal niveau. Deze zogenaamde proeftuinen fungeren als blauwdrukken en inspiratie voor de rest van de gezondheidszorg. Ook in Nederland staat het verbeteren van de financiële houdbaarheid van de gezondheidszorg hoog op de politieke agenda. Diverse regionale initiatieven zijn daarom aangewezen als proeftuinen om de gezondheid van de bevolking en de zorgkwaliteit te verbeteren en de groei van de zorguitgaven te beperken (Triple Aim). Een van de strategieën die in deze proeftuinen wordt gebruikt is het leveren van meer geïntegreerde zorg en het verhogen van de efficiëntie van de zorg door medisch gespecialiseerde zorg geleverd in de tweede lijn te verschuiven naar de eerste lijn, als een vorm van substitutie van zorg. Anderhalvelijnszorg (in het Engels: Primary Care Plus) geïmplementeerd in de proeftuin 'Blauwe Zorg' in de regio Maastricht-Heuvelland is een voorbeeld van een substitutie initiatief dat zich richt op medisch specialistische consulten in een eerstelijnssetting. In dezelfde periode werd het zorgpad 'Beter bewegen bij artrose' geïmplementeerd in proeftuin 'Anders Beter' gelegen in de Westelijke Mijnstreek. Het zorgpad richt zich op het veranderen van de aanvraag van artrose-gerelateerde diagnostiek door huisartsen en het verwijsgedrag van huisartsen naar de orthopeed. Dit is gedaan doormiddel van het stimuleren van een stepped-care aanpak op basis van richtlijnen om zo het gebruik van bestaande conservatieve behandelopties te optimaliseren en het efficiënte gebruik van diagnostische beeldvorming bij patiënten met/of een verdenking op knie- of heup artrose te verbeteren. Ondanks de verschillende aanpak van deze twee substitutie-initiatieven in de twee proeftuinen, zijn ze beide gericht op het voorkomen en vermijden van onnodige zorg en onnodige doorverwijzing naar de tweede lijn.

Hoofdstuk 1 introduceert het onderwerp van dit proefschrift. Het hoofdstuk geeft achtergrondinformatie over relevante zorgvraagstukken zoals fragmentatie van zorg en stijgende zorgkosten. Daarnaast gaat het in op verschillende strategieën om deze problemen aan te pakken, zoals geïntegreerde- en patiëntgerichte zorg. Verder beschrijft dit hoofdstuk de ontwikkeling van nieuwe zorgmodellen als antwoord op deze problematiek, waarbij de focus ligt op de Nederlandse substitutie-initiatieven anderhalvelijnszorg en het zorgpad 'Beter bewegen bij artrose'. Vervolgens zijn de doelen van dit proefschrift beschreven, namelijk: (1) het bestuderen van het effect van beide initiatieven op het verwijzingsgedrag naar de tweede lijn en/of de aanvraag

van diagnostische beeldvorming en de invloed van voorspellende kenmerken op de beslissing om al dan niet naar de tweede lijn te verwijzen, en (2) het onderzoeken van het effect van anderhalvelijnszorg op de gezondheidsgerelateerde kwaliteit van leven van de patiënt, de ervaren kwaliteit van zorg, de kosteneffectiviteit en het zorgvolume op regionaal niveau.

Hoofdstuk 2 onderzoekt de verwijsbeslissing na dermatologische zorg in de anderhalve lijn en de invloed van voorspellende patiënt- en consultkenmerken op deze beslissing. In deze retrospectieve studie zijn klinische gegevens gebruikt van patiënten die tussen januari 2015 en maart 2017 dermatologische zorg ontvingen in de anderhalve lijn. De verwijsbeslissing na anderhalvelijnszorg (d.w.z. een verwijzing terug naar de huisarts of een verwijzing naar de tweede lijn) was de primaire uitkomst. Stapsgewijze logistische regressiemodellen werden gebruikt om variaties in de verwijsbeslissing na anderhalvelijnszorg te beschrijven, met de leeftijd en het geslacht van de patiënt, het aantal consulten in de anderhalve lijn, de diagnose van de patiënt en de behandeld specialist als voorspellende factoren. Uit de resultaten bleek dat 80,2% van de patiënten die de anderhalve lijn bezochten voor dermatologische zorg werd terugverwezen naar de huisarts. Daarom lijkt dermatologische zorg geschikt te zijn voor anderhalvelijnszorg en kan hiermee het aantal doorverwijzingen naar de tweede lijn worden verminderd. Bovendien lieten de resultaten zien dat zowel de behandelend specialist als de diagnose van de patiënt de verwijsbeslissing na dermatologische zorg in de anderhalve lijn beïnvloeden. Met betrekking tot de diagnose werd een indicatie gegeven van geschikte diagnoses voor anderhalvelijnszorg. Maligne dermatosen lijken bijvoorbeeld minder geschikt voor de anderhalve lijn in verband met het grote aantal patiënten dat na anderhalvelijnszorg naar het ziekenhuis wordt verwezen. Aan de andere kant lijken diagnoses zoals haar- en nagelaandoeningen, die na anderhalvelijnszorg een lage doorverwijzing naar de tweede lijn hebben, bijzonder geschikt. Met deze indicaties dient echter voorzichtig te worden omgegaan, omdat klachten en symptomen van patiënten uiteindelijk kunnen leiden tot meerdere diagnoses, die niet altijd vooraf bij de huisarts bekend zijn. Over het algemeen kunnen de resultaten van deze studie worden gebruikt om de specialist- en patiëntprofielen voor de anderhalve lijn te verfijnen om zo de effectiviteit van anderhalvelijnszorg verder te optimaliseren.

Hoofdstuk 3 beschrijft de evaluatie van de verwijsbeslissing na orthopedische zorg in de anderhalve lijn en in het bijzonder de evaluatie van de invloed van diagnostiek op deze beslissing. Voor deze studie is gebruik gemaakt van retrospectieve monitoringgegevens

van patiënten die de anderhalve lijn bezochten voor orthopedische zorg tussen januari 2015 en december 2017. De primaire uitkomstmaat was de verwijsbeslissing na anderhalvelijnszorg (d.w.z. een verwijzing terug naar de huisarts of een verwijzing naar de tweede lijn). Onafhankelijke variabelen waren consult- en patiëntgerelateerde voorspellers. Om variaties in de verwijsbeslissing te beschrijven is gebruik gemaakt van logistische regressiemodellen. In lijn met de resultaten van het onderzoek naar dermatologische zorg in de anderhalve lijn, toonde ook deze studie aan dat zowel de behandelend specialist als de diagnose van de patiënt een significante invloed hebben op de verwijsbeslissing na anderhalvelijnszorg. Om het effect van de mogelijkheid om vanuit de anderhalve lijn diagnostiek (zoals echo's en MRI's) aan te vragen te onderzoeken werden de verwijsbeslissing en consultatie- en patiëntgerelateerde factoren vergeleken tussen twee periodes: P1 (toen het nog niet mogelijk was om aanvullende diagnostiek vanuit de anderhalve lijn aan te vragen) en P2 (toen de mogelijkheid om aanvullende diagnostiek aan te vragen werd geïntroduceerd). De resultaten tonen aan dat de mogelijkheid om aanvullende diagnostiek aan te vragen voor orthopeden die in de anderhalve lijn werken het aantal verwijzingen naar de tweede lijn aanzienlijk verminderde. Ondanks de aanzienlijke impact van de mogelijkheid om aanvullende diagnostiek aan te vragen, is het belangrijk om te bespreken in hoeverre de beschikbaarheid van diagnostiek past binnen de visie van anderhalvelijnszorg. Daarnaast is het selecteren van geschikte profielen voor specialisten en patiënten in de anderhalve lijn noodzakelijk om de effectiviteit en kosten van de zorg verder te optimaliseren.

Hoofdstuk 4 presenteert de resultaten van het onderzoek naar het zorgpad 'Beter bewegen bij artrose' dat is geïmplementeerd in de proeftuin 'Anders Beter'. Het doel van het zorgpad is om aan de hand van een 'stepped-care' aanpak het aantal aanvragen voor beeldvormende diagnostiek vanuit de eerste lijn te verminderen en om het aantal verwijzingen vanuit huisartsen naar orthopedische zorg in de tweede lijn terug te dringen. In 2015 is het zorgpad geïmplementeerd met behulp van educatieve bijeenkomsten, het verspreiden van richtlijnen en het opnemen van reminders in de verwijsapplicatie van huisartsen. Om het effect van het zorgpad op het aantal aanvragen voor beeldvormende diagnostiek in de eerste lijn en het aantal verwijzingen naar de orthopeed te evalueren, zijn heup- en kniegerelateerde zorgverzekeringsdeclaraties gebruikt in combinatie met declaraties voor andere gewrichten. Daarnaast is het aantal declaraties in de interventie regio vergeleken met het aantal declaraties in een controleregio. Om de interactie tussen regio (interventie en controle) en periode (pre- en postimplementatie) te onderzoeken is gebruik gemaakt van binaire

logistische regressieanalyses. Daarnaast is aan de hand van een willekeurige steekproef van patiëntendossiers informatie over de praktische toepassing van het zorgpad en het aantal heup- of knieprothesen toegevoegd. De resultaten laten zien dat de implementatie van het zorgpad heeft geleid tot een significante afname van beeldvormende eerstelijnsdiagnostiek van de knie in de interventieregio. Er werd echter geen significante afname gevonden van beeldvormende eerstelijnsdiagnostiek van de heup en, het aantal initiële heup- en kniegerelateerde consulten bij een orthopeed en van kans op een gewrichtsvervangende operatie. Een punt van aandacht voor toekomstige interventies en onderzoek is het verwijsgedrag van huisartsen naar de orthopeed, aangezien er juist een toename (in plaats van een gewenste afname) van het aantal initiële orthopedische consulten werd gevonden. Daarnaast is het focussen op de zorg voor heup- en knieartrose over de gehele breedte van het zorgspectrum van belang. Verder benadrukt dit onderzoek dat beeldvormende eerstelijnsdiagnostiek niet altijd nodig is om (in dit geval) artrose te diagnosticeren. Dit staat lijnrecht op de resultaten van het onderzoek naar orthopedische zorg in de anderhalve lijn waarin medisch specialisten juist benadrukten dat de aanwezigheid van diagnostiek een vereiste is om patiënten goed te kunnen diagnosticeren.

Hoofdstuk 5 laat zien of de gezondheid van de populatie en de ervaren kwaliteit van zorg door patiënten in de anderhalve lijn behouden blijven. Hiervoor zijn de gezondheidsgerelateerde kwaliteit van leven en de ervaren kwaliteit van zorg vanuit het perspectief van de patiënt vergeleken tussen patiënten die werden verwezen naar anderhalvelijnszorg en naar reguliere ziekenhuiszorg. In deze cohortstudie werden patiënten met een consult in de anderhalve lijn (interventiegroep) of tweede lijn (controlegroep) tussen december 2014 en april 2018 geïncludeerd. Met patiëntenvragenlijsten (afgenomen op T0, T1 en T2) werden de gezondheidsgerelateerde kwaliteit van leven en de ervaren kwaliteit van zorg gemeten. Propensity score matching werd gebruikt om te corrigeren voor mogelijke selectiebias. Vervolgens werden de interventie- en controlegroep vergeleken met lineaire modellen en Pearson chikwadraattesten. De resultaten laten zien dat de gezondheid en kwaliteit van zorg behouden blijven in de anderhalve lijn. Significante verschillen tussen de interventie- en de controlegroep werden enkel gevonden met betrekking tot de reistijd. Deze studie concludeert dan ook dat toekomstig onderzoek naar anderhalvelijnszorg zich meer zou moeten richten op kostengerelateerde uitkomsten.

Hoofdstuk 6 presenteert een vergelijking van de Kostenutiliteitsanalyse (KUA) en de Multicriteria-analyse (MCA) om de toepasbaarheid en geschiktheid van beide methoden

als ondersteuning van lokale besluitvormers bij het bepalen van de bredere waarde van nieuwe zorgmodellen te onderzoeken. Door anderhalvelijnszorg als case study te gebruiken, is onderzocht of het gebruik van een MCA in plaats van traditionele KUA de beslissing om te investeren in anderhalvelijnszorg verandert. Gegevens van patiënten die werden verwezen naar anderhalvelijnszorg of reguliere ziekenhuiszorg, werden verzameld via vragenlijsten en administratieve databases met een follow-up van drie maanden. Propensity score matching in combinatie met lineaire regressiemodellen is gebruikt om te corrigeren voor confounding. Om de onzekerheid in de resultaten te onderzoeken werden univariate en probabilistische sensitiviteitsanalyses uitgevoerd. Ondanks dat de resultaten van beide methodes laten zien dat anderhalvelijnszorg het dominante alternatief is, zijn er ook verschillen tussen de methodes waargenomen. Zo leverde de MCA aanvullend bewijs dat anderhalvelijnszorg leidt tot een verbeterde toegang tot zorg, een verbetering van gezondheidsgerelateerde kwaliteit van leven (met name door de component 'psychologisch welzijn'), en werd het benodigde budget om anderhalvelijnszorg effectief en betaalbaar te maken geschat op €521,42 per patiënt. Bovendien bleek de MCA minder gevoelig te zijn voor de gebruikte utiliteitsscores. Doordat de MCA aanvullende informatie verstrekt over een breder scala aan resultaten en over de betaalbaarheid van zorgmodellen kan de MCA een controleerbare en transparante evaluatie van nieuwe zorgmodellen vergemakkelijken. Echter, aandacht dient te worden besteed aan het vergroten van de bruikbaarheid van de MCA onder lokale besluitvormers.

Hoofdstuk 7 beschrijft óf en hóe anderhalvelijnszorg de regionale zorgvolumes op populatieniveau beïnvloedt. Eerder onderzoek beschrijft mogelijke mechanismen waarmee anderhalvelijnszorg een vervanging of aanvulling zou kunnen zijn op andere gezondheidsdiensten. Daarom is in deze studie gebruikgemaakt van retrospectieve zorgverzekeringsdeclaraties van de dominante zorgverzekeraar in de regio om de regionale zorgvolumes van 2015 tot 2018 te bepalen en deze te vergelijken met de landelijke trend. De resultaten tonen een toename van het totale volume laag-complexe medisch specialistische zorg in de interventieregio waaruit blijkt dat anderhalvelijnszorg er niet volledig in slaagt om substitutie van zorg te bewerkstelligen. Daarom is de conclusie dat naast het meten van de effecten van interventies op individueel patiëntniveau, ook de effecten op het totale zorggebruik op populatieniveau gemeten moeten worden. Bovendien is er tijd nodig om dit soort initiatieven door continue monitoring en evaluatie en daaruit volgende aanpassingen effectief te laten zijn. Daarom is een longitudinale dynamische evaluatiebenadering nodig om beleidsmakers te informeren.

Ten slotte wordt een samenvatting gegeven van de belangrijkste bevindingen van dit proefschrift, gevolgd door een reflectie op de theoretische en methodologische overwegingen. Verder zijn implicaties voor beleid, praktijk en onderzoek besproken.

Impact

Toenemende kosten in de zorg vormen een bedreiging voor de toegankelijkheid, kwaliteit en betaalbaarheid en daarmee de duurzaamheid van onze zorg. Als reactie hierop zijn in 2013 regionale samenwerkingsverbanden aangewezen door de toenmalige minister van Volksgezondheid, Welzijn en Sport als proeftuinen 'Betere zorg met minder kosten'. Binnen deze samenwerkingsverbanden werken regionale organisaties, zoals zorgverzekeraars, zorgaanbieders en patiëntenorganisaties, samen aan het opzetten van interventies met als doel betere gezondheid en betere patiëntervaringen tegen lagere kosten (de Triple Aim). In dit proefschrift staan de substitutie-initiatieven anderhalvelijnszorg (in het Engels: Primary Care Plus) geïmplementeerd in de proeftuin 'Blauwe Zorg' en het zorgpad 'Beter bewegen bij artrose' geïmplementeerd in de proeftuin 'Anders Beter' centraal.

ONDERZOEK

In het eerste deel van dit proefschrift wordt beschreven hoe de implementatie van anderhalvelijnszorg en het zorgpad invloed hebben op het verwijsgedrag naar de tweede lijn en op de aanvraag van diagnostische beeldvorming. De studies naar het effect van dermatologie en orthopedie in de anderhalve lijn laten zien dat een groot deel van de patiënten na een consult in de anderhalve lijn terug wordt verwezen naar de huisarts. Verder laten de resultaten zien dat zowel de behandelend specialist als de diagnose de verwijsbeslissing na een consult in de anderhalve lijn significant beïnvloeden. Met behulp van deze resultaten kunnen de patiënten- en specialistenprofielen voor anderhalvelijnszorg aangescherpt worden om het initiatief verder te optimaliseren. Daarnaast is aangetoond dat de mogelijkheid om diagnostiek aan te vragen vanuit de anderhalve lijn de verwijsbeslissing beïnvloedt, waarbij minder patiënten doorverwezen worden naar de tweede lijn wanneer aanvullende diagnostiek aangevraagd kan worden in de anderhalve lijn. Hiertegenover staan de resultaten van het onderzoek naar het zorgpad in de proeftuin 'Anders Beter'. Deze resultaten laten zien dat doormiddel van educatieve bijeenkomsten, het verspreiden van richtlijnen en het inbouwen van reminders in de verwijsapplicatie van huisartsen het aantal knie-gerelateerde diagnostiekaanvragen vanuit de eerste lijn verminderd kan worden. Met deze studie wordt benadrukt dat, in dit geval artrose-gerelateerde, diagnostische beeldvorming in de eerste lijn niet altijd noodzakelijk is om een diagnose te stellen.

Het tweede deel van het proefschrift richt zich op het anderhalvelijns initiatief in de proeftuin 'Blauwe Zorg' en beschrijft het effect op de uitkomsten van de Triple Aim en het regionale zorgvolume. Aan de hand van de onderzoeksresultaten kan worden geconcludeerd dat zorg in de anderhalve lijn tot dezelfde verbetering op het gebied van ervaren gezondheid leidt als reguliere ziekenhuiszorg. Daarnaast is aangetoond dat patiënten positief zijn over de kwaliteit van zorg in de anderhalvelijnscentra. Verder zijn de gemiddelde zorgkosten (met een follow-up van drie maanden) voor een patiënt die verwezen is naar anderhalvelijnszorg lager dan de zorgkosten voor een patiënt die verwezen is naar reguliere ziekenhuiszorg. Wat betreft de methode om de kosteneffectiviteit van initiatieven zoals anderhalvelijnszorg te evalueren en om te komen tot een beslissing om wel of niet te investeren in nieuwe zorgmodellen, blijkt de Multicriteria-analyse (MCA) een goede aanvulling te zijn op de meer traditionele Kostenutiliteitsanalyse (KUA)). Met behulp van de MCA kan informatie over een breder scala aan uitkomsten gegenereerd worden die bijdragen aan een controleerbaar en transparant besluitvormingsproces op lokaal niveau. Tenslotte is in dit proefschrift aangetoond dat ondanks de positieve effecten van anderhalvelijnszorg op de uitkomsten op patiëntniveau, dit niet automatisch leidt tot substitutie van zorg op regionaal niveau. Aan de hand van verzekeringsdata is aangetoond dat anderhalvelijnszorg tot een toename in het totale volume van laagcomplexe zorg in de regio Maastricht-Heuvelland heeft geleid. Om te komen tot succesvolle substitutie is dus ook aandacht nodig voor het regionale niveau en dienen substitutie-initiatieven ook op dit niveau gemonitord en geëvalueerd te worden.

RELEVANTIE

De resultaten van dit proefschrift dragen bij aan de wetenschappelijke kennis over de diverse initiatieven die wereldwijd worden ingezet om te komen tot substitutie van zorg. Deze substitutie-initiatieven kennen verschillende verschijningsvormen en onderzoek laat over het algemeen positieve resultaten zien met betrekking tot de tevredenheid van patiënten en de kwaliteit van zorg. De bevindingen over de zorgkosten zijn wisselend en veelal is er een gebrek aan robuuste economische evaluaties van dit soort initiatieven met een brede focus. In dit proefschrift is een dergelijke evaluatie wel aanwezig, waarbij naast de focus op individuele uitkomsten gerelateerd aan de Triple Aim ook gekeken is naar de invloed van anderhalvelijnszorg op populatieniveau.

Naast de wetenschappelijk relevantie dragen de resultaten van dit proefschrift ook bij aan het maatschappelijke vraagstuk over de financiële houdbaarheid en de duurzame

inrichting van het huidige zorgsysteem in Nederland (en in veel andere landen). De komende jaren zullen de zorguitgaven sneller stijgen dan onze economische groei kan bijbenen. Dit betekent dat er relatief steeds meer geld wordt uitgegeven aan zorg. Op landelijk niveau gaat dit uiteindelijk ten koste van andere belangrijke uitgaven, zoals onderwijs en veiligheid. Maar ook op individueel niveau gaan mensen dit voelen in hun portemonnee. De resultaten van dit proefschrift dragen bij aan de ontwikkeling en bijsturing van substitutie-initiatieven in de zorg die zich richten op het besparen van zorgkosten. Wetenschappelijke onderbouwing van de effecten van dit soort initiatieven wordt steeds belangrijker gevonden en dient bij voorkeur door een onafhankelijke partij te worden uitgevoerd. Door de verscheidenheid aan initiatieven die veelal op lokaal niveau worden ontwikkeld en geïmplementeerd is het voor zorgaanbieders en zorgverleners, zorgverzekeraars en beleidsmakers in andere regio's (binnen en buiten Nederland) interessant om de effecten van de initiatieven goed in beeld te hebben. Op deze manier kunnen nieuwe initiatieven ontwikkeld worden die verder bouwen op de resultaten van de bestaande initiatieven. Binnen de proeftuinen 'Blauwe Zorg' en 'Anders Beter' bieden de resultaten van dit proefschrift aanknopingspunten voor de verdere ontwikkeling en optimalisatie van de huidige substitutie-initiatieven om uiteindelijk daadwerkelijk te komen tot substitutie van zorg.

DOELGROEPEN

De onderzoeksresultaten van dit proefschrift zijn relevant voor patiënten, zorgaanbieders, zorgverleners, verzekeraars en beleidsmakers. Allereerst zijn de substitutie-initiatieven die in dit proefschrift aan bod komen gericht op het verbeteren van de zorg voor patiënten, waarbij het verbeteren van de gezondheid, het verbeteren van de ervaren kwaliteit van zorg en het verlagen van de zorgkosten centraal staan. De centrale rol van patiënten komt tevens terug in de betrokkenheid van de patiënten bij het evaluatieproces. Naast dat de patiëntenorganisatie Burgerkracht Limburg een belangrijke en betrokken partij is in beide proeftuinen, zijn patiënten ook direct betrokken bij de evaluatie van de initiatieven. Middels vragenlijsten zijn uitkomsten op het gebied van gezondheid en kwaliteit van zorg vanuit het patiëntenperspectief meegenomen. Daarnaast is het standpunt van patiënten bevraagd in kwalitatief onderzoek. Hierdoor levert dit proefschrift vanuit het patiëntenperspectief relevante inzichten op.

Andere betrokken initiatiefnemers in de twee proeftuinen die centraal staan in dit proefschrift zijn zorgaanbieders, zorgverleners, verzekeraars en beleidmakers. Naast

het opzetten van de initiatieven zijn deze partijen gedurende het evaluatieproces steeds betrokken geweest en vond continue informatie-uitwisseling plaats tussen de onderzoekers en de belanghebbenden. Aan de hand van deze informatie hebben de initiatiefnemers de substitutie-initiatieven waar nodig aangepast en verbeterd. Daarnaast hebben de onderzoeksresultaten geleid tot belangrijke aanbevelingen en randvoorwaardes voor het ontwikkelen van substitutie-initiatieven. Deze informatie is ook voor zorgaanbieders, zorgverleners, verzekeraars en beleidmakers buiten de proeftuinen interessant. Aangezien de substitutie-initiatieven positieve resultaten laten zien op zowel het gebied van het voorkómen van verwijzingen naar de tweede lijn en het aanvragen van diagnostiek als op de uitkomsten van de Triple Aim is het denkbaar dat andere regio's gelijksoortige initiatieven willen implementeren.

ACTIVITEITEN

Zoals al eerder genoemd is, zijn de initiatiefnemers van de substitutie-initiatieven in zowel de proeftuin 'Blauwe Zorg' als in de proeftuin 'Anders Beter' steeds actief betrokken geweest gedurende het evaluatieproces. Op regelmatige basis vond terugkoppeling plaats en werden tussentijdse inzichten en onderzoeksresultaten gedeeld. Daarnaast werd ook samen met de belanghebbenden gereflecteerd op de onderzoeksresultaten om deze te verifiëren en om zo bij te dragen aan een betere interpretatie van de resultaten. Deze samenwerking heeft veel kennis opgeleverd. Naast dat er op regelmatige basis overleg plaatsvond met de belanghebbenden, zijn de onderzoeksresultaten ook nog op andere manieren verspreid. Zo zijn de hoofdstukken van dit proefschrift gepubliceerd in internationale wetenschappelijke tijdschriften dan wel onder review van een tijdschrift. Daarnaast zijn er ook twee verschillende Nederlandstalige rapporten verschenen rondom de monitoring en evaluatie van de substitutie-initiatieven en over de doorontwikkeling van anderhalvelijnszorg in de proeftuin 'Blauwe Zorg'. Deze rapporten maken de onderzoeksresultaten en de geleerde lessen toegankelijk voor de belanghebbenden en andere geïnteresseerden. In deze rapporten worden naast de bevindingen van de substitutie-initiatieven ook de onderzoeksmethoden beschreven. Verder zijn de onderzoeksresultaten gepresenteerd op wetenschappelijke congressen in binnen- en buitenland. Vanuit de Academische Werkplaats Duurzame Zorg zijn daarnaast diverse symposia georganiseerd om de onderzoeksresultaten te delen met belanghebbenden en andere geïnteresseerden uit de regio Limburg en de rest van Nederland. Dit bood tevens de mogelijkheid voor proeftuinen om onderling kennis uit te wisselen en zo van elkaar te leren.

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About
THE AUTHOR

Esther van den Bogaart was born on March 21, 1990 in Someren, the Netherlands. After finishing secondary education at the Varendonck College in Asten, she studied Health Sciences at Maastricht University, obtaining her bachelor's degree in 2011. In 2012, Esther obtained her Master of Science in Health Education and Promotion at the University of Maastricht. Her thesis was about motives and considerations about preimplantation genetic diagnosis for couples with hereditary breast cancer. After obtaining her master degree, Esther worked as an administrative assistant for a home care organisation and an insurance company and in the meantime she travelled for four months. In 2014, Esther started working as a researcher of the Home Sweet Home study, at CIRO in Horn.

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SCIENTIFIC PUBLICATIONS

Scientific articles in international journals

van den Bogaart EHA, Spreeuwenberg MD, Kroese, MEAL, van Hoof SJM, Hameleers N, Ruwaard D. Patients' perspectives on a new delivery model in primary care: A propensity score matched analysis of patient-reported outcomes in a Dutch cohort study. Journal of Evaluation Clinical Practice. 2021;27(2): 344.

van den Bogaart EHA, Spreeuwenberg MD, Kroese, MEAL, van den Boogaart MW, Boymans TAEJ, Ruwaard D. Referral decisions and its predictors related to orthopaedic care. A retrospective study in a novel primary care setting. PLOS ONE 2020;15(1).

van den Bogaart EHA, Kroese, MEAL, Spreeuwenberg MD, Martens H, Steijlen PM, Ruwaard D. Reorganising dermatology care: predictors of the substitution of secondary care with primary care. BMC Health Services Research. 2020;20(1):510.

van den Bogaart EHA, Kroese, MEAL, Spreeuwenberg MD, Ottenheijm RPG, Deckers P, Ruwaard D. Does the implementation of a care pathway for patients with hip or knee osteoarthritis lead to fewer diagnostic imaging and referrals by general practitioners? A pre-post-implementation study of claims data. BMC Family Practice. 2019;20(1):154.

Submitted articles

van den Bogaart EHA, Spreeuwenberg MD, Kroese, MEAL, Ruwaard D. The effect of a new intervention in between primary and secondary care on regional healthcare volumes: substitution or addition?

van den Bogaart EHA, Kroese, MEAL, Spreeuwenberg MD, Ruwaard D, Tsiachristas A. Economic evaluation of new models of care: does the decision change between Cost-Utility Analysis and Multi-Criteria Decision Analysis? (Accepted for publication in Value in Health)

Reports

van den Bogaart E, Quanjel T, Kroese M, Spreeuwenberg M, Hameleers N, Westra D, Ruwaard D. Monitoring en evaluatie van substitutie van zorg in drie Limburgse proeftuinen. Evaluatierapport. 2019. GIldeprint.

Kroese M, van den Bogaart E, Ruwaard D. Verkenning mogelijkheden doorontwikkeling Anderhalvelijnszorg Maastricht-Heuvelland. Rapport. 2020.

Conference contributions

van den Bogaart E, Kroese M, Spreeuwenberg M, Hameleers N, Ruwaard D. Anderhalvelijnszorg: wat zijn de effecten op kosten en volumes van zorg? NHG-Wetenschapsdag, 5 February 2021, the Netherlands. (Presentation)

van den Bogaart E, Kroese M, Spreeuwenberg M, Ruwaard D, Tsiachristas. Economic Evaluation of New Models of Care: Does the Decision Change between Cost-Utility Analysis and MULTI-Criteria Decision Analysis? Virtual ISPOR Europe 2020, November 2020. (Poster presentation)

Ruwaard D, Quanjel T, van den Bogaart E, Westra D, Hameleers N, Kroese M. Evaluating Primary Care Plus Interventions in the Netherlands: Discussion of Methods and Results regarding the Quadruple Aim. 19th International Conference of Integrated Care, 3 April 2019, San Sebastian, Spain. (Workshop)

van den Bogaart E, Spreeuwenberg M, Kroese M, van den Boogaart M, Boymans T, Ruwaard D. Is orthopaedic care suitable for substitution of outpatient hospital care with primary care? A retrospective analysis of referral data. European Health Management Association annual conference 2018, 21 June 2018, Budapest, Hungary. (Presentation)

van den Bogaart E, Kroese M, Spreeuwenberg M, Martens H, Steijlen P, Ruwaard D. Reorganising dermatology care: predictors to substitute secondary care with primary care. 18th International Conference of Integrated Care, 24 May 2018, Utrecht, the Netherlands. (Oral poster presentation)

van Hoof S, van den Bogaart, Quanjel T, Westra D. Toekomstmuziek in Anderhalvelijnszorg. Symposium, 28 March 2018, Maastricht, the Netherlands. (Presentation)

Ruwaard D, van den Bogaart E, Quanjel T, Kroese M, Spreeuwenberg M. Het monitoren van anderhalvelijnszorg in de drie Limburgse proeftuinen. Werkconferentie Substitutie, 26 September 2016, Maastricht, the Netherlands. (Presentation)