PAYMENT AND ECONOMIC EVALUATION OF INTEGRATED CARE

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Payment and Economic Evaluation of Integrated Care

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To Elli

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CHAPTER 1

General introduction

1.1 Health and social burden of chronic diseases

The rapid growth and ageing of global population accompanied by unhealthy health behaviour (i.e. smoking, physical inactivity, unhealthy diet and harmful use of alcohol) lead to increasing prevalence of chronic diseases. Worldwide, non-communicable diseases (NCDs) cause more than 63% of all deaths, a number that is expected to increase by 15% at 2020 and by more than 20% at 2030. These mortality rates affect females and males almost equally [1]. Many non-communicable diseases are chronic diseases, like cardiovascular diseases (48% of total deaths due to NCD), cancers (21%), chronic respiratory diseases (12%), and diabetes (4%), which are on the top list of the deadliest NCDs. Twenty five percent of the deaths from NCD occur before the age of 60 [1]. Chronic diseases have high impact on global morbidity too. Seventy seven percent of total disability adjusted life years (DALYs) are caused by chronic diseases; mostly by cardiovascular diseases (23% of total DALYs), neuropsychiatric conditions (20%), cancers (11%), respiratory diseases (5%), and diabetes (2%) [2,3].

Furthermore, chronic diseases disproportionately affect people at lower socioeconomic status. At patient-level, chronic diseases and poverty create a vicious cycle whereby poverty exposes people to behavioural and environmental risk factors for chronic diseases and, in turn, the resulting chronic diseases may become an important driver to the downward spiral that leads families towards poverty. At country level, 80% of deaths related to NCDs occur in low-middle income countries [4]. Furthermore, the social impact of chronic diseases is even larger taking into account that they influence the mortality and quality of life of their informal care givers and family [5,6].

1.2 Economic burden of chronic diseases

It is estimated that over the next 20 years, chronic diseases will cost more than 48% of the global GDP in 2010 [7]. They account for a large extent of the increasing health care expenditure [8] because their direct and indirect costs in health care are a sizeable share of a country's GDP. This share is continuously increasing due to the rising prevalence of chronic diseases [9]. As a result, chronic diseases constitute a great challenge to economies and a threat to the sustainability of health care systems worldwide.

This challenge is even greater when considering that chronic diseases have long-term macroeconomic impact on consumption, capital accumulation, labour productivity and labour supply [2]. Consequently, chronic diseases jeopardize global economic growth worldwide. The negative macroeconomic consequences are more severe in developing countries for two reasons [7]. The first reason is because these countries have a higher need for human and financial resources to maintain their relatively high economic

growth compared with higher income countries. The second reason is that in developing countries citizens frequently do not have a universal health insurance coverage and therefore, chronic diseases entrench people in poverty due to catastrophic expenditures for treatment.

1.3 Integrated care and disease management programs

A large percentage of chronic diseases is preventable through the reduction of four main behavioural risk factors: tobacco use, physical inactivity, harmful use of alcohol and unhealthy diet. If these risk factors were eliminated, around 75% of heart disease, stroke and type 2 diabetes as well as 40% of cancer would be prevented [1,10]. This potential to improve the sustainability of health care systems urges the switch of care for people with chronic diseases from acute and reactive to preventive and proactive. In other words, care for people with chronic diseases requires an integrated, multidisciplinary package of well-coordinated care that includes (besides prevention) monitoring and active participation of patients [11]. However, health care systems are currently focusing on acute, mono-disciplinary and segmented care. Therefore, many health care authorities worldwide strive to redesign chronic care.

Integrated care is the most promising concept in redesigning care to tackle the increasing threat of chronic diseases. It refers to a "range of approaches deployed to increase coordination, cooperation, continuity, collaboration, and networking across different components of health service delivery" [12]. It puts the patient and his or her individual needs in the centre and organizes care around the patient, thereby seeking to reduce redundancies and fragmentation in health care delivery [13]. However, the term integrate care is vague and confusing because there is no commonly agreed definition and because different terms (such as coordinated care, managed care, seamless care) have been used interchangeably with integrated care [14]. The underlying denominator of integrated care definitions is the improvement of outcomes for population groups with diverse and complex needs. This is reflected in the World Health Organization (WHO) definition of integrated care, which is described as "a concept bringing together inputs, delivery, management and organization of services related to diagnosis, treatment, care, rehabilitation and health promotion. Integration is a means to improve services in relation to access, quality, user satisfaction and efficiency" [15]. Kodner and Spreeuwenberg (2002) also addressed that the aim of integrated care is "to enhance quality of care and quality of life, consumer satisfaction and system efficiency for patients with complex, long-term problems cutting across multiple services, providers and settings" [14]. Busse et al. (2008) stated that integrated care aims to: (1) improve quality of care delivery, (2) ensure professional adherence to disease specific protocols and guidelines, (3) reduce

unnecessary hospital utilization by strengthening the primary care sector, (4) share financial responsibility with other stakeholders, and in the long-term, (5) contain the increasing chronic care expenditure [16].

Many governments around the world have experimented with models for integrating care for chronic disease. The first integrated care models were developed in the United States in the 1980s. The Kaiser Permanente's "pyramid of care" model in California and the "Evercare" model in Minnesota are the most well-known and influential ones [11]. The American experience with the identification of chronic disease and providing care according to the patients' needs have been influential in Europe and elsewhere [17-19]. It has been the basis of the introduction of case management in all Primary Care Trusts in the United Kingdom National Health System in 2007, which aimed to improve the quality and accessibility of care for people with chronic conditions and contain associated costs. In Sweden, many county councils offer chains of care for diabetes, dementia, and rheumatoid arthritis [20]. In France, the formation of local provider networks for ambulatory patients was stimulated through the 2002 Patients' Rights and Quality of Care Act [21]. Likewise, in the province of Ontario in Canada networks of family doctors and local health integration networks were formed [22].

However, the most wide-spread model towards integrating care for chronic disease was the implementation of disease management programs (DMPs) [23]. Schrijvers (2009) defines these programs as 'a group of coherent interventions designed to prevent or manage one or more chronic conditions using a systematic, multidisciplinary approach and potentially employing multiple treatment modalities' [24]. In many cases, the disease management programs are based on Wagner's Chronic Care Model (CCM), which provides a framework of elements that must be considered when developing improvement strategies for providing care for people with chronic diseases, originally including: (a) self-management support, (b) decision support, (c) delivery system design, (d) clinical information systems, (e) health care organization, and (f) community resources and policies [25]. The CCM was later extended to put more emphasis on patient safety, care coordination and case management [11]. Several studies concluded that the CCM improves patient care and health outcomes [26,27].

The first structured DMPs were developed in the United States in the 1990s and were offered by Health Maintenance Organizations (HMOs), Medicare, Medicaid and other payers to treat a wide variety of chronic disease such as diabetes, congestive heart failure and asthma. In Germany large numbers of patients with a chronic disease were enrolled in DMPs that were introduced on a large scale [2]. In the Netherlands, recently developed DMPs [28] built further upon earlier successful shared care or "transmural" care projects [19,29,30] supported by a strong primary care sector [31]. DMPs were also promoted as part of the health care reforms in Austria [32,33] and France [23] to address the needs of people with chronic diseases.

Literature reviews concluded that DMPs improve quality of care [34,35], patient's quality of life and physical exercise [36] , patient self-management [37], and reduces hospitalization [38,39]. However, there is large variation in the components of provided DMPs and the participating population which causes large variation in their effectiveness [40].

1.4 Payment schemes

Payment schemes are key-factors in influencing stakeholder behaviour, and can thus be used to stimulate their collaboration and steer health care delivery systems toward integration [13]. This is because they are not only influenced by their intrinsic motivation to provide good quality care but also by financial motives [15]. Traditional health care provider payment schemes include salary, capitation, and fee-for-service (FFS) [2]. These payment schemes lack specific incentives to stimulate multidisciplinary collaboration and improve the quality of chronic care [13,41]. A salary fails to stimulate integration of care because there are potential incentives to accept only healthy patients (cream skimming) and to refer complex cases to more costly secondary services (dumping). Capitation provides caregivers with an incentive to spend a little amount of time on each patient such that more patients can be enrolled that generate compensation. As such, chronically ill are financially unattractive as they require more time and services to treat, at the expense of the physician, who would otherwise receive the same remuneration for treating a healthier patient who merely requires an occasional simple, quick treatment. FFS, on the other hand, generates an incentive to provide as many refundable services as possible. While FFS reduces the incentive to avoid the chronically ill, there is little incentive for caregivers to provide high quality of care and adequately address the needs of patients with chronic diseases. [16]. Consequently, these traditional payment schemes are unable to facilitate integration and high quality of chronic care [2].

Adequate financial incentives are prerequisites to improve chronic care [28]. Therefore, several European countries have implemented different payment schemes in order to provide financial incentives to health care providers for implementing integrated care [42]. Similar to the US [43], adaptations of pay-for-coordination (PFC) (e.g. Austria, Denmark, France), pay-for-performance (PFP) (e.g. England, France), bundled payments (e.g. the Netherlands), and global payments (e.g. Germany) targeting different stakeholders (mainly health care providers and payers) have been introduced the last decade in Europe [16].

Evidence from the literature suggests that PFP can potentially be effective [44], bundled payment may reduce health care utilization [45], and global payment may improve quality and reduce health care expenditure [46]. However, there is lack of

comprehensive information regarding which and how payment schemes have been implemented and robust evidence about how the organizational structure, quality, and efficiency of chronic care was impacted as a result.

1.5 Price setting for care provided by DMPs

Setting a price for the provision of a DMP is not a single task. Apparently, apart from the interventions provided by a DMP, two major cost components should be adequately anticipated and incorporated in this price in order to ensure the financial viability of DMP providers. The first component is the case-mix of patients included in a DMP. For instance in the Netherlands, health care insurers purchase chronic care from DMP providers based on a predefined price for an expected case-mix of chronic patients. This price is negotiable between the two parties; it is paid on a yearly basis per patient included in a DMP and it is roughly risk-adjusted [47]. Failing to correctly anticipate the case-mix of patients could lead to a negotiated price lower than the actual costs. The second component includes the development and implementation costs of DMPs. The development costs include training costs, ICT costs, and costs of redesigning the care delivery process, while the implementation costs include multidisciplinary team meetings, the costs of coordination between care givers, the costs of monitoring and feedback. To which extent these costs are included in the payment of a DMP provider is often unclear. This is despite recommendations to report these costs separately from the health care utilization costs and to include them in the price of implementing a DMP [48].

The variability in health care utilization costs based on the case-mix of patients included in DMPs as well as the development and implementation costs of DMPs are poorly investigated so far.

1.6 Economic evaluation of integrated care

Although the implementation of integrated care programs and more specifically of DMPs is growing, the evidence about their cost-effectiveness is still inconclusive [2,35,49,50]. Some studies have found that DMPs are cost-saving [51-54] or cost-neutral [55]. Other studies however, have found positive net costs but still cost-effective results [56-60] or hardly any indications of cost-effectiveness [61].

This can largely be explained by the variation in study design, outcome measures, and costing methods used in the economic evaluations of DMPs [57,62]. Furthermore, the definition of DMPs is subject to national or local health care settings because they are very context-specific and setting-dependent, which might be another reason for dif-

ferences in their outcomes so far. What contributes to the discrepancies in the economic evaluations of DMPs however, is the lack of a methodological framework to facilitate such evaluation [50]. Many sophisticated frameworks and complicated techniques have been developed in conventional cost-effectiveness analysis the last decades. However, their adequacy for evaluating DMPs is limited because of the multi-faceted nature of these programs, their context-dependent implementation, and the broad spectrum of outcomes related to their implementation. The lack of a consistent framework for evaluating DMPs has made comparisons of results hardly possible, meaning that much time and financial resources have been spent inefficiently [63]. Current decision-making incorporates traditional cost-effectiveness studies that may not be suitable for the comparison between DMPs and usual care. This is because DMPs are complex, multifaceted interventions that have a great variety of effects such as improved self-management capabilities, changed disease perceptions, coordination and continuity of care, reduced risk factors and complication rates, improved health-related quality of life, improved wellbeing, reduced burden on informal caregivers, etc. These cannot be expressed in a single unit of effect like a quality-adjusted life year (QALY) that is traditionally used in economic evaluations of health care interventions. Therefore, the establishment of a methodological framework to perform an analysis incorporating the most relevant costs and effects is desirable in performing economic evaluations of DMPs valuable to decision making.

Previous studies that attempted to develop such frameworks [63,64] have acknowledged the fact that this would be methodologically challenging [48,50]. Such a framework could provide decision makers with information about the consequences of integrated care in quality of care, patient health, and monetary terms and therefore, support them in understanding whether current expenses could be regarded as investments [65]. However, a thorough analysis based on an evaluation framework that incorporates all related costs and effects and covers all relevant aspects of integrated care is still to be found.

1.7 Thesis aims and research questions

The aims of this thesis were to: 1) investigate payment schemes implemented to promote integrated care and assess their impact, 2) explore the variability in health care utilization costs determined by the case-mix of patients involved in DMPs as well as to examine the variability in the development and implementation costs of DMPs, and 3) determine the costs and effects of DMPs and develop a framework to facilitate a thorough economic evaluation of DMPs.

The following research questions were formulated to address each of these aims.

Addressing aim 1:

1. Which payment schemes have been implemented in Europe to promote integration of chronic care, what were the facilitators and barriers to their implementation, and how did stakeholders perceive their success?
2. How was the new bundled payment system in the Netherlands designed and implemented and what were the facilitators and barriers to introducing this scheme?
3. What was the impact of different financial agreements to promote integrated care in Europe on national health care expenditures?

Addressing aim 2:

4. Which factors on patient-level and organizational-level are associated with the costs of patients with different chronic diseases enrolled in a DMP?
5. What is the variability in development and implementation costs among various DMPs and what are its determinants?

Addressing aim 3:

6. What are the changes in costs and outcomes after the implementation of DMPs, what are their determinants, and what is their short-term (i.e. 1-year) cost-effectiveness?
7. What is the long-term (i.e. 2-year) cost-effectiveness of implemented DMPs?
8. What would be an adequate methodological framework to facilitate a systematic and broader economic evaluation of DMPs including the most relevant outcomes and cost categories?

1.8 Thesis outline

Each research question is answered in a separate chapter in this thesis. The chapters are categorized into three parts, reflecting the three aims. Part 1 concerns payment schemes that promote integrated care and highlights their impact. In particular, Chapter 2 investigates different payments schemes implemented in Europe to promote integrated care and their perceived impact on different aspects. A more detailed description of such a payment scheme, i.e. the bundled payment scheme which was introduced in the Netherlands in 2010, is provided in Chapter 3. In Chapter 4 we empirically investigate the impact of these payment schemes on health care expenditure. Part 2 explores the variability and determinants of health care utilization costs of DMP enrolees (Chapter 5) and development and implementation costs of DMPs (Chapter 6). The last part of the thesis (i.e. Part 3) provides empirical evidence about the changes in costs and effects after the implementation of DMPs (Chapter 7), including a cost-effectiveness analysis of the most comprehensive DMPs over a 2-year period (Chapter 8). It also presents a methodological framework to systematically and thoroughly evaluate DMPs (Chapter 9). The answers to the research questions are summarised in the general discussion of the

thesis together with recommendations for future research (Chapter 10). Finally, Chapter 11 provides a summary of the thesis in English and in Dutch as well as information about the author.

PART A

Payment of integrated care

CHAPTER 2

Exploring payment schemes used to promote integrated chronic care in Europe

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Abstract

The rising burden of chronic conditions has led several European countries to reform healthcare payment schemes. This paper aimed to explore the adoption and success of payment schemes that promote integration of chronic care in European countries. A literature review was used to identify European countries that employed pay-for-coordination (PFC), pay-for-performance (PFP), and bundled payment schemes. Existing evidence from the literature was supplemented with fifteen interviews with chronic care experts in these countries to obtain detailed information regarding the payment schemes, facilitators and barriers to their implementation, and their perceived success.

Austria, France, England, the Netherlands, and Germany have implemented payment schemes that were specifically designed to promote the integration of chronic care. Prominent factors facilitating implementation included stakeholder cooperation, adequate financial incentives for stakeholders, and flexible task allocation among different care provider disciplines. Common barriers to implementation included misaligned incentives across stakeholders and gaming. The implemented payment schemes targeted different stakeholders (e.g. individual caregivers, multidisciplinary organizations of caregivers, regions, insurers) in different countries depending on the structure and financing of each health care system. All payment reforms appeared to have changed the structure of chronic care delivery. PFC, as it was implemented in Austria, France and Germany, was perceived to be the most successful in increasing collaboration within and across healthcare sectors, whereas PFP, as it was implemented in England and France, was perceived most successful in improving other indicators of the quality of the care process. Interviewees stated that the impact of the payment reforms on healthcare expenditures remained questionable.

The success of a payment scheme depends on the details of the specific implementation in a particular country, but a combination of the schemes may overcome the barriers of each individual scheme.

2.1 Introduction

Chronic conditions place a largely increasing economic burden on national healthcare budgets worldwide because of their rising incidence and prevalence [[66]. Traditional healthcare payment schemes are designed for predominantly acute care settings and are therefore, restricted in their ability to tackle inefficiencies present in chronic care [16]. Tackling these inefficiencies could potentially reduce the increasing economic burden of chronic conditions [2].

Integrated chronic care refers to a "range of approaches deployed to increase coordination, cooperation, continuity, collaboration, and networking across different components of health service delivery" [12].It puts the patient and his or her individual needs in the center and organizes care around the patient, thereby seeking to reduce redundancies and fragmentation in healthcare delivery [13]. Specifically, integrated chronic care aims to: (1) improve quality of care delivery, (2) ensure professional adherence to disease specific protocols and guidelines, (3) reduce unnecessary hospital utilization by strengthening the primary care sector, (4) share financial responsibility with other stakeholders, and in the long-term, (5) contain the increasing chronic care expenditure [16].

Several countries have experimented with innovative approaches to achieve integration of chronic care [2]. Wagner's Chronic Care Model (CCM) is one of the most influential approaches and is based on the notion that productive interactions between stakeholders results in higher quality chronic care [25]. The CCM was used in many European countries to design and implement Disease Management Programs (DMPs) to achieve integration of chronic care [67]. DMPs are defined by the Disease Management Association of America as "a system of coordinated healthcare interventions and communications for populations with conditions in which patients self-care efforts are significant". The success of DMPs is largely dependent on the financing context and payment mechanisms relevant to the various stakeholders involved, as they are not only influenced by their intrinsic motivation to provide good quality care but also by financial motives [15].

Payment schemes are key-factors in influencing stakeholder behavior, and can thus be used to stimulate their collaboration and steer healthcare delivery systems toward integration [13]. [16][16]Therefore, several European countries have implemented different payment schemes in order to implement integrated healthcare delivery systems with regard to chronic care. However, there remains in the literature a lack of comprehensive information regarding which and how payment schemes have been implemented as well as about how the organizational structure, quality, and efficiency of chronic care was impacted as a result.

The aim of this paper is to provide an overview of payment schemes that have been implemented in Europe to promote integration of chronic care, highlight the facilitators and barriers to their implementation, and assess how stakeholders perceived their success.

2.2 Methods

A literature review was conducted to identify payment schemes introduced to improve the integration of chronic care in European countries since 1997. We searched in Google Scholar and Pubmed for relevant published papers using combinations of the following keywords: "chronic care", "financing", "payment", "integrated", "coordinated", and "disease management". The references of the resulting papers were then scanned to find additional publications relevant to our study. For our initial selection of countries, we searched websites of governmental organizations and research institutes as well as in conference proceedings to collect additional information. Based on this information, we made the final selection of those countries that had implemented payment schemes to improve chronic care on a national level (policies on local or regional level were excluded). Payment schemes of interest excluded traditional caregiver payment schemes (e.g. fee-for-service (FFS), capitation, salary), which are not particularly targeted at disease management, coordination of healthcare delivery and ultimately integration of chronic care [16]. From the literature we obtained information about the implemented payment scheme(s) in each country, the financial incentives provided, the barriers and facilitators for their implementation, and their impact on chronic care delivery and expenditure.

To supplement the findings from the literature, we conducted telephone interviews in the countries of interest. Potential interviewees were experts in chronic care (payment schemes) and were identified from the literature (authorship), (non-) governmental agencies (contact persons), and conference programs (presenters). Literature was also used to develop a template for the interviews. The template incorporated elements previous studies have considered while investigating similar, related topics [2,31,68][28]. Interviewees had broad, first-hand knowledge of the payment schemes in question, and ranged in expertise including researchers, health insurers, and patient organizations. The interview template consisted of two consecutive parts. Part one consisted of semi-open questions addressing the policy aim, details pertaining to stakeholders and policy implementation, and changes and realizations since implementation. Questions addressed the most relevant policy, and in some cases, multiple relevant policies. Part two consisted of statements relating to the success of implementation, effect on integration of care, effect on financing scheme, and policy evaluation. Response options for these statements had a five-point likert scale ranging from for example strongly

disagree to strongly agree (Appendix 1). The interviews were held in English or Dutch, were transcribed in English, and were analyzed qualitatively.

2.3 Payment schemes for integrated chronic care

Traditional healthcare payment schemes include salary, capitation, and FFS [16]. They are not specifically designed to stimulate integrated care or improve the quality of chronic care [13,41,69].

Providing adequate financial incentives is a key instrument in achieving the implementation of integrated chronic care [13], as they influence stakeholder behavior [25,69]. Several countries have implemented alternative payment schemes with financial incentives that overcome the limitations of traditional payment schemes to promote integrated chronic care [16,69]. These payment schemes include: pay-for-coordination (PFC), pay-for-performance (PFP), and bundled payment. The theoretical foundations of these payment schemes are summarized in Table 1.

PFC consists of payments to one or more providers to coordinate care between certain care services [70,71]. It seeks to provide an incentive for the extra effort required by stakeholders to cooperate with one another, share organized, transparent information on healthcare delivery and health outcomes, often set to predefined standards. As a result, PFC is expected to control unnecessary utilization, promote provider integration as well as encourage continuity of care. Its implementation is considered as feasible with relatively little effort.

PFP is a direct payment to a health care provider for achieving defined and measurable goals related to improvements in the process and/or outcomes of chronic care delivery [71,72]. PFP seeks to improve the quality of care by generating additional compensation for caregivers that deliver high quality of care and comply with guidelines. Its implementation may be more or less demanding, depending on the level of ICT and the number and type of quality indicators. However, it weakly promotes integration between healthcare providers.

Bundled payment is a single payment for all multidisciplinary care required by a patient for one particular chronic disease during a predefined period of time [43,71,73]. It aims to control unnecessary health care utilization, encourage high quality of care, and promote integration between health care providers. Its implementation faces the challenge of defining the content of the care bundles and determining a price per bundle. Bundled payment provides a direct incentive to health care providers to increase their profit margin by reducing inefficiencies. It may be attractive to payers because they run relatively little financial risk.

Table 1 Theoretical foundations of payment schemes that facilitate integration of care

	Increasing aggregation of services into a unit of payment		
	Pay-for-coordination	**Pay-for-performance (physician)**	**Bundled Payment**
Description	Payments to providers providing care coordination services that integrate care between providers	Physicians receive differential payments for meeting or missing performance benchmarks	A single "bundled" payment, which may include multiple providers in multiple care settings, is made for services delivered during an episode of care related to medical condition or procedure.
Attributes			
Population			
Episode of care		√	√
Multiple types of delivery organizations			√
Fee for newly specified services	√		
Objectives			
Control unnecessary utilization	√√		√√
Encourage high quality	√	√√	√√
Promote provider integration	√√	√	√√
Operational feasibility	√√	√√	√√
Financial Incentives			
Patient level			
Provider Level	Payment for support services not covered under a FFS or capitation	Higher payment when goals are achieved	Increase profit margin by reducing inefficiencies
Payer Level	Avoidance of unnecessary and/or inefficient care (e.g. double payment for same treatment)		Limits financial exposure

Based on: Mechanic and Altman 2009; Schneider et al., 2011; adjusted by the authors; Note: PFC and PFP have the same aggregation level of services into a unit of payment

2.4 Results

From the literature review, five countries were identified as having implemented payment schemes to promote the integration of chronic care on a national level. These in-

cluded Austria, England, France, Germany, and the Netherlands. Most payment schemes implemented in these countries were adaptations and a specific operationalization of the payment schemes described in the previous section. These adaptations were necessary because several of the payment schemes for integrated care were developed originally in the United States and had to be transferred to the European context. In many cases, the payment schemes were accompanied by restructuring of chronic care financing.

The next sections explore the implementation of various policies using PFC, PFP, and bundled payment schemes as a means to promote the integration of chronic care, including a description of the policies, when and by whom they were implemented, as well as which incentives they provide for various stakeholders (where applicable), their barriers and facilitators, and their perceived impact. The findings from literature and interviews are presented below together. The citation to each interview is given with the anonymous codes in brackets. A list of interviewees, their anonymous codes, and their profession is provided in Appendix 2.

2.5 Introduced payment schemes

PFC schemes were evident in Austria, France, and Germany, PFP in England and France, and bundled payment in the Netherlands. A summary of the financial incentives provided per payment scheme can be found in Table 2.

2.5.1.1 Pay-for-coordination

In Austria, the Health Reform Act of 2005 was implemented by the Ministry of Health to promote integration and coordination of care, improve efficiency, resource allocation and funding by pooling financial resources and promoting DMPs [32,32,33]. This reform created financial pools at state level by combining 1–2% of the budget of social health insurers with that of regional governments. These pooled funds were available for integrated care projects between primary and secondary care [74]. This was expected to overcome segmentation between the social health insurance scheme to fund outpatient care and the provincial health funds in inpatient care and to be economically beneficial for both schemes (A.B.). The 2005 health reform act also promoted DMPs, funded by social health insurance, targeting general practitioners (GPs) and promoting their engagement in the coordination of integrated care efforts. On a national level, a DMP has only been implemented for diabetes, incorporating guidelines for cardiovascular risk assessment [75]. This was accompanied by a PFC payment scheme as physicians received an initial premium (€53) upon patient enrolment in DMP and a quarterly payment (€25) to supplement the traditional FFS. GPs qualified for providing DMPs if they participated

Table 2 Financial incentives for integrating chronic care in each country

	Patient Level	Provider Level	Pooler/Payer Level
Austria		• €53 initial + €25 quarterly per patient enrolled in DMP	• 1-2% of the combined existing budgets across sectors to be designated for integrated chronic care projects
France	• Reduced copayment if patient enrols in DMP	• €40 annual per patient enrolled in DMP (PFC) • 0% to 30% annual bonus (PFP)	
England		• 0% to 30% annual bonus (PFP)	
The Netherlands		• Price negotiated between insurer and care group (bundled); Performance is a factor in price negotiations	
Germany	• Reduced copayment if patient enrols in DMP • Additional services (e.g. self-management education) only reimbursable if patients participate in DMPs	• €75 per patient per year for coordination costs (PFC) • Additional remuneration for disease specific education programs provided within a DMP • 1% of ambulatory budget and 1% of hospital budget was earmarked for integrated care projects	• €153 annual per patient enrolled in DMP for coordination costs (PFC) • Remuneration for enrolling chronically ill based on morbidity and mortality indicators

PFC: pay-for-coordination; PFP: pay-for-performance; DMP: disease management program

in a basic training regarding care coordination, and attended refreshment courses. Additional courses on patient education were optional, for which physicians would receive an additional remuneration (A.B.). In Austria, there is no choice of insurer or competition among health insurance funds, as insurance is mandatory and contingent on place of residence or employer [33].

In France, the Health Insurance Reform Act (2004) was an initiative targeting the primary care sector, promoting the expanded use of DMPs for 30 chronic diseases including diabetes, COPD, cardiovascular diseases, muscoskeletal diseases and certain cancers [68] (F.C.). Initiated as a negotiation between the social health insurance and the association of GPs (F.A.), the aim of this program was to improve quality of care, patient monitoring, promote continuous medical education to communicate common guidelines to care providers, alleviate financial burden associated with unnecessary procedures, and strengthen the role of the GP [12]. It was accompanied by a PFC payment scheme as GPs received supplemental D 40 for care coordination [68] (F.A.). Patients benefited from waived co-payments, reduced waiting times as well as self-education and training programs. GPs were not obligated to engage in DMPs. While patients were

free to supplement social health insurance with private health insurance, they are not free to choose the insurer within the social health insurance scheme (F.A.).

In Germany, the Risk Structure Compensation Reform Act was introduced in 2002. Under this scheme, health insurers received a fixed fee per patient per year for costs in primary and secondary care [76]. This compensation aimed on one hand to avoid cream-skimming from the insurers at the expense of chronically ill patients (G.B.) and on the other hand to promote DMPs, which were believed to improve quality of chronic care [74,77,78]. Initially, DMPs existed for breast cancer, diabetes, coronary heart disease, asthma, and COPD and these were extended to more disease areas [68]. Health care providers negotiated collectively for which conditions to provide DMPs and had uniform documentation forms for all patients independent of health insurer [77]. To recruit DMP participants, the insurer could reduce or waive patients' co-payments [78,79]. The remuneration was contingent on whether the services provided were in line with the disease specific DMP guidelines (G.C.) [80]. Concerning the PFC payment, the reform introduced financial incentives for health insurers and health care providers. Health insurers who enrolled chronically ill patients in DMPs were provided with D 85 per patient per year and coordinating physicians received D 75 per patient per year for coordination costs, including necessary documentation [68,79]. Providers also received additional payment for disease specific education programs for registered patients. In 2009, when the participation in a DMP was no longer used as adjustor in the risk-equalization formula, health insurers received €180 per patient per year for coordination costs which was decreased by 2012 to €153 [81] (G.A.). In addition, health insurers could retain 1% of the ambulatory budget and 1% from the hospital budget and make them available for integrated care projects. As a result, health care providers had a strong incentive to develop integrated care projects (extending to primary and secondary care) because they risked losing a share of their budgets to competitors [82]. The Social Health Insurance Competition Strengthening Act was implemented in 2007 to further strengthen and promote care integration [68,74,83]. It extended the one-percent start-up provision for integrated care contracts until 2008, moved to include long-term care in integrated care contracts, and allowed non-medical healthcare professionals to contract with insurers. Long term integrated care contracts shared the aim of DMPs, but differed in that they were funded partially by the aforementioned start-up provision. Furthermore, integrated care contracts focused on coordination between hospitals and rehabilitation practices, most often addressing orthopedic indications (i.e. hip and knee surgery) [83] (G.C.).

2.5.1.2 Pay-for-performance

In France, Contrats d'amelioration des pratiques individuelles2 (CAPI) was launched as a voluntary pilot in 2009 and expanded in 2012 [84] (F.B.). CAPI were signed by GPs

on voluntary basis for three years and provided addition remuneration on top of their FFS income. These contracts set a PFP payment scheme in which GPs were rewarded financially, not for specific disease treatments but rather for adequately registered patient records and for following evidence based guidelines. The number of performance indicators started at 16 and increased to 29 (F.C). GPs could possibly earn an additional €6,000 annually (30% of their base salary) when they achieved over 85% of the targets and treated more than 1,200 patients [84].

In England, the Quality and Outcomes Framework (QOF) was introduced in 2004 [85]. The QOF offered PFP contracts to GPs, by which GPs were rewarded additionally based on 146 performance indicators within four domains; clinical standards, organizational standards, patient experience, and additional services [86-88]. This aimed to enhance the quality of primary care provided according to national guidelines, and its implementation was justified by the success of various quality-improvement initiatives that had been introduced since 1991 [85]. In 2006, adjustments were made to the system, altering minimum and maximum payment thresholds, dropping, modifying, and introducing new indicators [88]. In 2009/2010, further adjustments were made, adding new indicators for heart failure, chronic kidney disease, depression, and diabetes, removing two indicators from the patient experience domain, and adjusting the point values of several indicators [33]. Initially, £1.8 billion was designated to reward GPs by a possible 25% salary increase, which was later increased to 30% [88]. Exception reports, through which GPs can decide to exclude patients from the calculation of certain irrelevant performance indicators, ensured a focus on relevant and appropriate targets [87-89]. Patients can use information, published by the NHS information center, to compare and choose a GP practice in which to enroll [88].

2.5.1.3 Bundled payment

In the Netherlands, a bundled payment was piloted in 2007 with diabetes and expanded in 2010 to include COPD and cardiovascular disease management [47,89-91]. The aim of these payment reforms was to improve coordination between providers, promote the use of DMPs, strengthen adherence to medical guidelines, and increase quality of patient records [91]. Under the new payment scheme, chronic care is coordinated by groups of providers (called care groups) that implement DMPs organized in integrated centers in primary care or in groups of cooperating general practices, paramedical care givers and/or hospitals [47]. Insurers negotiated with care groups a predefined fee (bundled payment) that covered all care needed by a patient with a particular chronic disease for a year (excluding inpatient care, medication, medical devices, and diagnostics). Then care groups negotiate with and subcontract individual care providers for the care delivery [91] (N.B.; N.A.). Negotiations generate significant price variations between care groups for a particular group of patients i.e. different prices for different diabetes DMPs, serving

to promote competition-induced quality improvements, on the basis of, but not limited to, performance measures, which are described in national care standards [47,92] (N.B.). Insurers are free to choose whether they contract care groups based on the bundled payment system, or instead provide care groups only with an additional payment for the organization, coordination, and transparency of care, while continuing to reimburse individual providers on a FFS basis. Patients are free to choose their GP and can change insurance company annually, choosing the most relevant, but least costly package to suit their medical needs [91](N.A., N.B.).

2.5.2 Facilitators and barriers per payment scheme

While each payment scheme was unique, they often experienced similar facilitators and barriers to their adoption and implementation. The most frequent facilitators were adequate financial incentives, flexible work roles (i.e. enabling nurses and GPs to share duties and responsibilities), and stakeholder cooperation, while the most frequent barriers were misaligned incentives across stakeholders (e.g. the FFS of a dietician is higher than the share of the bundled payment that they receive) and gaming (e.g. enrolling pre-diabetic patients in diabetes DMP). Table 3 provides an overview of facilitators and barriers per payment scheme, which are explained in the following sections.

Table 3 Overview of facilitators and barriers per payment scheme

	Facilitators	Barriers
Pay-for-coordination	• Stakeholder cooperation (AUS, GER) • Patient Demand (AUS, FRA) • Adequate financial incentive for GPs (FRA)	• Gaming (GER) • Misaligned incentives between stakeholders (AUS) • GP Opposition (AUS,GER, FRA) • Virtual budget (AUS) • Inflexible task allocation (AUS)
Pay-for-performance	• Adequate financial incentive for GPs (ENG, FR)	• Gaming (ENG) • Defining performance indicators (ENG)
Bundled Payment	• Stakeholder cooperation (NL) • Flexible task allocation (NL)	• Gaming (NL) • Lack of transparency (NL) • Lack of comprehensive means to address multi-morbidity (NL)

AUS=Austria; ENG=England, FR=France; GER=Germany; NL=the Netherlands

2.5.2.1 Pay-for-coordination

The PFC scheme was facilitated by the: (a) cooperation between health insurers and healthcare providers (Austria) (A.A.), (b) cooperation between insurers and government (Germany) (G.B.), (c) patient demand for integrated care services, as a result of the increased awareness about its benefits (Austria, France) (A.A.;F.C.) and (d) adequate financial incentives for GPs to engage patients in DMPs (France) (F.C.).

However, the implementation of PFC initiatives experienced the following barriers. GP opposition in the implementation of PFC was evident because GPs (a) feared restrictions in their medical autonomy due to evidence-based guidelines (Austria, Germany, France) (A.A.;A.B.;G.A.;F.C.;F.A.), (b) rejected the notion of an education requirement to establish eligibility for DMP participation (Austria) (A.A.;A.B.), (c) considered PFC less financially attractive than FFS as they could earn more from the latter payment scheme in the same consultation time per patient (Austria) [74] (A.A.; A.B.), and (d) were not enthusiastic due to the additional administrative requirements associated with PFC (France) (F.A.). Moreover, misaligned incentives between health insurers and provinces in Austria (A.B.; A.A.) jeopardized the implementation of PFC.

Other barriers to PFC included the mislabeling of standard care procedures as integrated care to receive wrongly the PFC fee (Germany) (G.C.). Virtual budgets were a barrier in Austria because the decision to reallocate and merge a percentage of the GP and hospital budgets was left to the respective parties without providing a concrete incentive for them to do so (A.B.). Furthermore, inflexible task allocation between different providers (Austria) (A.B.) has also obscured the implementation of PFC schemes.

2.5.2.2 Pay-for-performance

The facilitation of PFP in England and France was attributed primarily to the strong financial incentives for GPs as they could increase their income by 30% (E.B.; F.C.). In England, the pre-existing strong collaborations in the primary sector facilitated administrative and documentation demands of the QOF [82,87] (E.B.). There is speculation in England as to whether physicians optimized their financial rewards by labeling failed targets as exception reports, but gaming in this sense is expected to be minimal, if evident at all [86] (E.B.). In addition, the definition of performance indicators was troublesome in England [86]. In France, there were no barriers identified in the implementation and actual uptake of the recently introduced CAPI program [84] (F.A.; F.C.), probably because it was introduced only recently.

2.5.2.3 Bundled payment

In the Netherlands, the bundled payment scheme was facilitated by a high level of commitment by policy makers, care providers, and health insurers as well as a flexible responsibility allocation and task delegation from GPs and specialists to nurse practitioners and GP assistants [91,92] (N.A.;N.B.). Barriers to the success of this scheme included: (a) care groups referred costly patients unnecessarily to hospitals in order to protect their budget [92] (N.B.), (b) lack of transparency in cost-pricing of bundled payments, stemming from underdeveloped IT systems and resulting in distrust between insurers and care groups, as insurers are sceptical about double payments (e.g. FFS and bundled

payment) for the same care provision [90,92,93], and (c) absence of a systematic way of addressing a patient with multi-morbidities [68,74,92].

2.5.3 Perceived impacts of integrated chronic care payment schemes

Table 4 provides an overview of the impact of integrated chronic care payment schemes implemented in Austria, France, England, the Netherlands, and Germany as perceived by the interviewees and supplemented with literature. In most cases, interviewees stated that the implementation of a payment scheme had a structural impact on the financing and process delivery of chronic care, while the perceived impact on decreasing the growth of chronic care expenditure was negative or sceptical at best. All but the PFC payment scheme in Germany were perceived as having introduced new budgetary constraints in the healthcare system. This implied that additional money was required by the healthcare system (without regarding or considering the possible return to investment) as a result of the payment scheme implementation. Detailed information regarding the perceived impacts of the various payment schemes is provided below.

Table 4 Perceived effects of integrated chronic care payment schemes

	PFC			PFP		Bundled
	AUS	FR	GER	ENG	FR	NL
Structural impact on financing and process delivery of chronic care	–	+	++	+	+	++
Increased provider cooperation within a care sector	+	+	–	?	+	+
New collaboration agreements between care sectors	+	?	–	–	–	?
Promotes integrated financing of different care sectors	+	?	–	–	–	+
Introduced new budgetary constraints on healthcare system	+	+	–	++	?	+
Decreased growth of chronic care expenditure	–	?	?	?	?	–

PFC= pay-for-coordination, PFP=pay-for-performance, AUS=Austria, ENG = England, FR =France, GER=Germany, NL=the Netherlands; A composite of interview responses was formed to characterise each payment scheme, per country, as follows: ++ =strongly agree; +=agree; ?=N/A or unknown; – = disagree; –– = strongly disagree

2.5.3.1 Pay-for-coordination

The implementation of PFC was perceived by the interviewees as successful with relatively high uptake in Germany and France while in Austria, it was perceived more troublesome, as actors did not respond to the incentives with which they were provided. In Austria, the uptake of the DMP implementation was low because GPs considered the imposed administrative burden high [74] as well as because care groups that applied

for funding integrated programs between primary and secondary care were established prior to, and independently of, financial pools reform (A.A).

Moreover, as the interviewees stated, PFC resulted in change toward enabling an improved financing structure for chronic care (Germany, France) (G.A.; G.B.; G.C.; F.C.), increased provider cooperation (Austria, France) (A.A.:F.B), introduced new collaboration agreements between care sectors (Austria,) (A.A.), promoted integrated financing of different care sectors (Austria) (A.A.; A.B.), and introduced budgetary constraints (A.A.:F.B). In Germany, interviewees stated that PFC did not promote provider cooperation, collaboration agreements between care sectors, and integrated financing of different care sectors (G.A.; G.B.; G.C.). These failures were also reported in the literature [82]. The perceived impact of PFC on the growth of chronic care expenditure was doubtful in France and Germany while, it was considered to be negative in Austria (A.A.).

2.5.3.2 Pay-for-performance

The uptake of PFP was 100% in England [88], 30% during its infant stage in France (F.A.; F.C.), which climbed to 90% within 3 years (F.A.; F.B.; F.C.) [86]. According to the interviewees, PFP led to positive structural changes in chronic care financing and chronic care delivery (England, France), and increased provider cooperation within primary care (France). However, in both countries it was not designed to lead to new collaboration agreements or promote integrated financing between primary and secondary care (E.A.; E.B; F.A.;F.B.; F.C.).

2.5.3.3 Bundled payment

In the Netherlands, the bundled payment scheme was perceived as having a positive structural impact on financing and process delivery of chronic care, increased provider cooperation within the primary care sector, and promoted the integration of financing of different care sectors (N.A.; N.B.). However, the interviewees stated that the bundled payment introduced new financial constraints in the health care system and failed to decrease the growth of health care expenditure up till now. It was also believed that it improved protocol adherence and record keeping, and promoted competition between care health care providers, [91,93] (N.A.; N.B.). According to the interviewees, the impact on new collaboration agreements between care sectors remained inconclusive.

2.6 Discussion

After providing an overview of payment schemes introduced in Austria, France, Germany, England, and the Netherlands, several discussion points come to light. First and foremost is that is some countries, the payment reforms were accompanied by financial

arrangements targeting different stakeholders. PFC was introduced together with the financial pooling in Austria and the risk structure compensation in Germany, provided financial incentives and means to financial poolers and payers. On the other hand, the implementation of PFP in France and England targeted the financial reward of primary care physicians only. In the Netherlands however, the implementation of bundled payment provided financial incentives to health insurers and health care providers. These differences imply that reforming payment schemes in chronic care depends strongly on the structure of a health care system. Therefore, financial incentives targeted to key stakeholders may enable the successful implementation of payment reforms.

Furthermore, amongst the countries explored, with the exception of England, PFC was originally implemented as the primary mechanism to achieve integration. Explicit integrated care programs, most commonly DMPs, are particularly appealing as they are specifically outlining and incentivizing responsibilities per stakeholder. GP opposition was a barrier to implementing PFC in Austria and France. This opposition was largely attributable to concerns about reduced medical autonomy, and increased educational and administrative requirements rather than the means of financing. Eventually, a clear-cut link between responsibility and reward appeared to mobilize stakeholders toward implementation. As a result, collaboration was stimulated between providers and across care sectors. This collaboration is necessary in achieving integrated chronic care delivery systems [68]. Considering that PFC was limited to increased collaboration, it becomes apparent that the addition or combination with other payment schemes would be more successful in attaining additional policy goals, such as reduced growth of chronic care expenditure.

Other combinations of payment schemes could include PFP with bundled payment, such that the amount of the case-mix payment fluctuates partially according to performance indicators. This combination might enhance quality of care by providing strong financial incentives to payers and/or health care providers. In the Netherlands, there is such an implicit combination, as performance indicators are taken into account when health insurers and care groups negotiate about the prices of the bundled care packages [88]. However, the use of performance indicators is limited without a concrete agreement on how exactly they determine the bundled payment [47,93].

The introduction of performance indicators in payment schemes must be encouraged cautiously because they might reduce the intrinsic motivation of health care providers to provide the utmost quality of care. Shortcuts and pitfalls of the PFP system are continuously being evaluated in England, and the indicators reconsidered to optimize desired results [44,94]. This reconsideration is currently manifesting itself as a gradual shift from process indicators to including and expanding relevant outcome indicators.

The suspicion of gaming and evidence of misaligned incentives in all payment schemes suggests a vulnerability of the healthcare system. As healthcare budgets be-

come rigid and stakeholders are increasingly responsible for their individual budget, it is inevitable that gaming might occur to secure and protect these budgets. Therefore, it is interesting to consider shared-savings schemes for aligning stakeholders to enhance integration of chronic care. There are many examples of shared-savings programs in the U.S. [71] to provide European decision makers with inspiration and experience toward experimenting with such schemes. In the Netherlands, there are currently pilots of payment schemes that incorporate shared-savings set-up by health insurers and health care providers in primary and secondary care [93]. However, they are still in an infant phase and no evidence about their impact is available.

The strengths of this study include the combination of literature and expert opinion to provide an overview of payment schemes implemented in European countries, explore the facilitators and barriers to their implementation, and discuss their perceived impact. However, it has several limitations. First, it includes a limited number of interviewees that precludes the generalizability of the findings regarding the perceived impact of each payment scheme. However, the interviewees were predominantly well-known researchers with hands-on experience with DMPs on European level. This could mitigate any biases in their perceptions about the impact of the payment schemes. Second, it discusses only the payment schemes strictly related to integrated chronic care, as other policies or the wider health care system in each country are not investigated. We acknowledge the relevance of these aspects to fully understand a payment scheme but such complicated issues cannot be addressed in the scope of this explorative paper. Our overview can be the base for further in-depth investigation of each payment scheme in each country. Third, all interviewees stated that the payment schemes had a structural impact on chronic care financing but their opinions did not converge about the decrease of healthcare expenditure growth after implementation. Therefore, we cannot draw a consistent conclusion on this issue from our results presenting a limitation of the qualitative character of this study. We are currently conducting quantitative research that focuses on the impact of the introduced payment schemes on health care expenditure.

2.7 Conclusions

Payment schemes are valuable tools in stimulating the integration of chronic care delivery. The development of such payment schemes in Europe targeted those stakeholders who were expected to adjust their behavior, and provided them with adequate financial incentives. All payment reforms appeared to have changed the structure of chronic care delivery. PFC, as it was implemented in Austria, France and Germany, was perceived to be the most successful in increasing collaboration within and across healthcare sectors,

whereas PFP, as it was implemented in England and France, was perceived most successful in improving other indicators of the quality of the care process. Interviewees stated that the impact of the payment reforms on healthcare expenditures remained questionable.

Our findings suggest that initiating collaborations in chronic care can be stimulated with PFC payments and further integration of care can be facilitated by adding other payment schemes such as bundled payments. Elements of performance based payments are definitely important for stimulating competition and improving quality of care. Other payment agreements, such as shared savings, should also be considered to overcome gaming and misaligned incentives between stakeholders. All this information can help decision makers to further improve the (re)design of payment schemes in Europe toward a blended payment scheme that facilitates integration of chronic care.

Appendix 1 Interview template

Interview Part 1:

Core Policy:	
Policy title	
Date	
What is the aim of the policy?	
What did this policy change with regard to financing and managing chronic conditions in *country name*?	
Who initiated this policy?	
What is the target audience of this policy?	
How widely implemented is this policy?	
Are there any plans to update/ alter this policy? Why & in which way?	
How do governmental agencies, healthcare providers, insurers, and/or patients view this policy?	
How successful do you believe this policy is up until now?	
How successful do you believe this policy will be in the future?	
Are there perceived barriers or obstacles to this policy functioning as intended?	
On the other hand, are there facilitating factors?	

Interview Part 2:

Please indicate below the degree to which you agree/disagree with the following statements, with regard to the core policy indicated in part 1. If you feel there is another core policy, we would greatly appreciate it if you could indicate which policy it is, and complete the following form a second time, for that policy as well.

Statements related to success of implementation:

1. The policy changed positively the financing structure of chronic care.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

2. The level of commitment of policy makers is high.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

3. The level of commitment of healthcare providers is high.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

4. The level of commitment of health insurers is high.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

5. The level of commitment of patients is high.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

6. The policy implementation was met with resistance.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

7. No additional training was required for the health care providers to implement the policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

8. The policy has had structural or systematic impact on the delivery and/or financing of chronic disease management.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

9. Previously implemented policies facilitated the implementation of this policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

10. The ICT systems of chronic care was a barrier for implementation.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

Statements related to Integration of Care:

1. The policy led to more cooperation between providers.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

2. As a result of policy implementation, chronic patients became more involved in their care.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

3. Chronically ill patients are free to choose whether they engage in policy provisions.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

4. As a result of policy implementation, new collaboration agreements have been made between care sectors (e.g. between primary, secondary, and/or home care).

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

5. For the coordination of chronic care, the integration of ICT systems of the care sectors involved increased quality in care provided.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

Financing Scheme:

1. The policy increases efficiency in chronic care.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

2. There are adequate financial incentives for the stakeholders (e.g. patients, providers, insurers) to participate/adopt the policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

3. The policy imposes budgetary constraints on the healthcare system.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

4. The policy promotes the integration of financing of different care sectors involved in chronic care.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

5. Risk selection of financially "unattractive" chronic patients to health insurers is decreased following the implementation of this policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

6. The growth of the chronic care expenditure decreased after the introduction of this policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

Policy Evaluation:

1. Ongoing national evaluation of the policy is lacking.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

2. Sufficient data is available to evaluate the impact of the policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

3. There is political interest regarding the impact of the policy.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

4. Public visibility/Transparency exists regarding policy impact.

Strongly Agree	Agree	Neutral	Disagree	Strongly Disagree
□	□	□	□	□

Appendix 2 List of interviewees

Country	Interviewee code	Gender	Interview date	Profession
Austria	A.A.	Female	4 May 2012	Researcher Health Economics and Health Policy
	A.B.	Male	23 May 2012	Researcher Health Economics and Health Policy
Denmark	D.A.	Male	4 April 2012	Researcher and Consultant Public Health
	D.B.	Female	25 April 2012	Researcher Health Services and Chronic Care Management
	D.C.	Female	2 May 2012	Researcher Integrated Care
France	F.A.	Female	15 April 2012	Researcher Health Economics
	F.B.	Male	18 April 2012	Researcher Health Economics and Health Services
	F.C.	Male	12 July 2012	Researcher and Consultant Health Economics and Policy
Germany	G.A.	Male	11 April 2012	Researcher Health Economics and Health Services
	G.B.	Male	10 May 2012	Director of Sickness Fund
	G.C.	Female	14 May 2012	Researcher Health Policy
Netherlands	N.A.	Male	14 May 2012	Manager in Patient Organization
	N.B.	Male	18 May 2012	Researcher Public Health
England	E.A.	Male	1 May 2012	Researcher Health Services Management
	E.B.	Female	9 July 2012	Researcher Health Policy and Financing Health Systems

CHAPTER 3

Towards integrated care for chronic conditions: Dutch policy developments to overcome the (financial) barriers

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Abstract

Chronic non-communicable diseases are a major threat to population health and have a major economic impact on health care systems. Worldwide, integrated chronic care delivery systems have been developed to tackle this challenge. In the Netherlands, the recently introduced integrated payment system – the chain-DTC – is seen as the cornerstone of a policy stimulating the development of a well-functioning integrated chronic care system.

The purpose of this paper is to describe the recent attempts in the Netherlands to stimulate the delivery of integrated chronic care, focusing specifically on the new integrated payment scheme and the barriers to introducing this scheme. We also highlight possible threats and identify necessary conditions to the success of the system. This paper is based on a combination of methods and sources including literature, government documents, personal communications and site visits to disease management programs (DMPs).

The most important conditions for the success of the new payment system are: complete care protocols describing both general (e.g. smoking cessation, physical activity) and disease-specific chronic care modules, coverage of all components of a DMP by basic health care insurance, adequate information systems that facilitate communication between caregivers, explicit links between the quality and the price of a DMP, expansion of the amount of specialized care included in the chain-DTC, inclusion of a multi-morbidity factor in the risk equalization formula of insurers, and thorough economic evaluation of DMPs.

3.1 Introduction

Chronic non-communicable diseases are a major threat to population health and an increasing challenge to health care. Worldwide, chronic conditions cause about 60% of all deaths [3], a number that is expected to increase by 17% in the next 10 years [10]. Parallel to the increasing prevalence of chronic diseases, there is growing recognition that chronic care delivery requires an integrated, multidisciplinary package of well-coordinated care that includes prevention, monitoring and maintenance treatment while empowering chronically ill patients to participate actively in their treatment [68]. Both the need to contain the costs of chronic care treatment and the recognition that currently, health care systems are ill-equipped to provide integrated care has led many health care authorities worldwide to redesign chronic care delivery.

During the 1980s, the first structured integrated care or disease management programs were developed in the United States. At that time, disease management was mainly seen as an instrument of cost controllers to reduce hospital (re)admissions and hospital days. During the 1990s the nature and scope of these programs was widened and Health Maintenance Organizations (HMOs), Medicare, Medicaid and other payers now offer a wide variety of disease management programs for conditions such as diabetes, congestive health failure and asthma. The American experience with the identification of chronic conditions and providing care according to the patients' needs have been influential in Europe and elsewhere [17-19]. It has been the basis of the introduction of case management in all Primary Care Trusts in the United Kingdom National Health System in 2007, which aimed to improve the quality and accessibility of care for people with chronic conditions and contain associated costs. At the same time many other European countries have experimented with various forms of integrated care delivery. In Germany large numbers of patients with a chronic disease were enrolled in disease management programs that were introduced on a large scale. Until 2009, there was a strong incentive to enroll patients because these programs were a separate component of the risk structure compensation schemes for sick funds. In turn, the sick funds provided strong incentives to doctors to enroll patients [2]. In Sweden, many county councils offer chains of care for diabetes, dementia, and rheumatoid arthritis [20]. In France, the formation of local provider networks for ambulatory patients was stimulated through the 2002 Patients' Rights and Quality of Care Act [21]. Likewise, in the province of Ontario in Canada networks of family doctors and local health integration networks were formed [22]. Most countries implement a spectrum of parallel policies targeting specific elements of the chronic care continuum (e.g. The Netherlands, France, Germany, Sweden, Canada) and some countries have developed a national vision on chronic disease control with nationwide integrated care strategies covering the entire care continuum from health promotion to care for complex chronic disease patients (e.g.

Australia, Denmark, England) [2,12]. There is no single approach that fits all countries, and new initiatives to provide integrated care need to be embedded in national and provincial structures for the delivery and financing of care [2,31].

As a result of these different approaches across the globe, many different definitions of disease management exist. A frequently cited definition is the definition of the Disease Management Association of America (DMAA), which defines a DMP as 'a system of coordinated health care interventions and communications for populations with conditions in which patient self-care efforts are significant' [67]. In most cases, the disease management programs are based on Wagner's Chronic Care Model (CCM), which provides a framework of elements that must be considered when developing improvement strategies for providing care for people with chronic diseases, originally including: (a) self-management support, (b) decision support, (c) delivery system design, (d) clinical information systems, (e) health care organization, and (f) community resources and policies [25]. The CCM was later extended to put more emphasis on patient safety, care coordination and case management [11].

In the Netherlands, recently developed disease management programs (DMPs) [28] built further upon earlier successful shared care or transmural care projects [19,29,95] supported by a strong primary care sector [31]. However, due to the lack of coordination between the Dutch policies to put the CCM components in place [96], true integration in the care for chronic diseases is still far from having been achieved in the Netherlands [31]. Therefore, further improvements are envisioned, especially in the areas of self-management support, clinical information systems, and decision support. As a result, the delivery systems for chronic care are currently undergoing a re-design, having the introduction of integrated payment system in 2010 as the cornerstone policy to develop a well-functioning and sustainable integrated care system for people with chronic conditions.

So far, there is little information available about the new payment system and its position in the spectrum of other Dutch policies to stimulate integrated chronic care. This information would enable patient representatives, insurers and care providers to better understand and prepare for their (potential) role in the transformation of chronic care. This information is also necessary for researchers studying the (cost-)effectiveness of disease management programs to position their findings in the policy context of chronic care. Therefore, the aim of this paper is to describe the recent attempts in the Netherlands to stimulate the delivery of integrated chronic care, focusing specifically on the new integrated payment scheme and the facilitators and barriers to introducing this scheme. A review of relevant literature and government documents is the first core research method adopted to achieve the aim of the paper. Keywords, such as 'integrated care', 'disease management programs', 'transmural care', 'chronic care', 'The Netherlands' were used to identify relevant literature, whereas the websites of governmental organizations and institutions involved or influencing chronic care were searched for relevant

information. The second core research method, that provides information directly from the work floor, is a series of ongoing personal communications with project leaders and care providers of 22 different DMPs that participate in a government program to stimulate disease management for diabetes, cardiovascular risk management, COPD, and anxiety/depression. In addition, we participated in workshops and symposia in the Netherlands, where stakeholders of chronic care discussed concerns for and advantages of the new developments in this field.

Following the introduction (Section 1), the current policies (Section 2) and barriers (Section 3) for achieving integrated care are outlined. By current policies we mean the policies up until 2009. Thereafter, we describe the integrated payment scheme that was introduced in 2010, including its intended incentives (Section 4), followed by its potential benefits (Section 5) and possible threats to its success (Section 6). We further discuss the necessary conditions for the new integrated payment scheme to work (Section 7) and the final section concludes with a call for evaluation (Section 8).

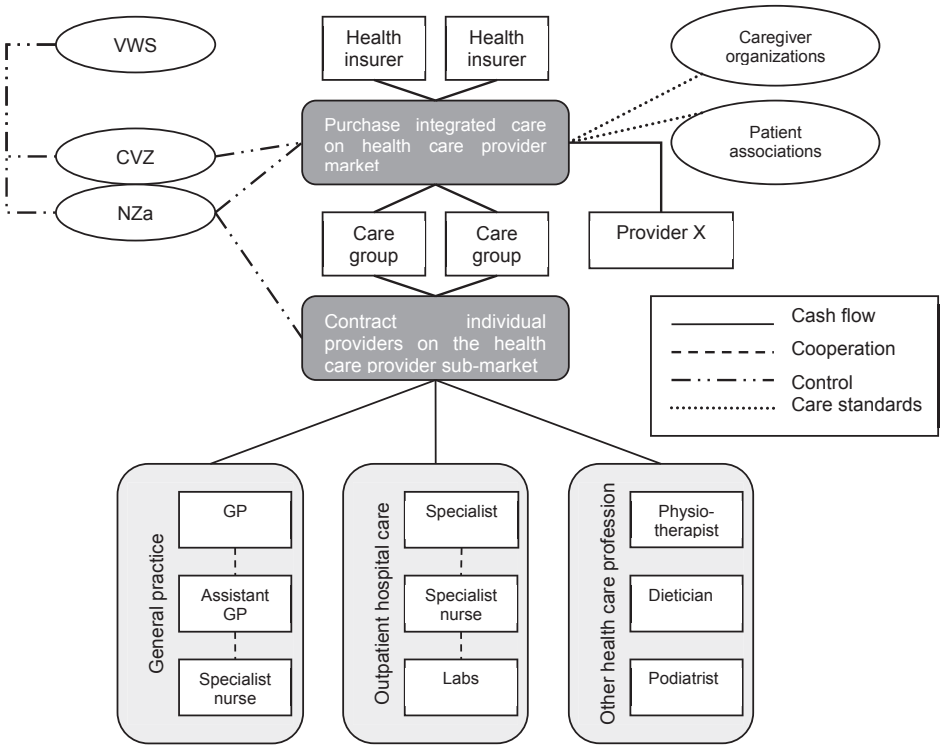
3.2 Current policy influencing integration of care

During the last two decades, Dutch authorities have aimed to increase efficiency in the health care system by introducing regulated competition [97,98]. To achieve that, a series of health care reforms was gradually undertaken towards a market oriented health care and a national health insurance system [97,98]. The most important reform was the introduction of the Health Insurance Act (Zvw) in 2006, which set the foundations for a regulated market in the Dutch health care system. The goal of this reform was to increase efficiency of health care provision by encouraging health insurers to act as purchasers on behalf of health care consumers. Consumers and patients are seen as important players who make rational choices between insurers, benefit packages and providers based on information about volume, price and quality [98]. Full implementation of this Act is still in progress [99].

3.2.1 Health care insurance

Under the Health Insurance Act, the distinction was abandoned between the compulsory public sickness funds for persons with less than a defined income, and private, mostly voluntary health insurance for private entrepreneurs and people with higher incomes [18,97]. Health insurers are now required to offer a standard package of basic health care insurance to every applicant, regardless of pre-existing condition. It is mandatory for every citizen to have this basic benefit package. In addition, the insurers offer voluntary supplementary insurance packages at additional cost. Patient choice is enhanced by allowing consumers to change health care insurer every year, thereby giving them

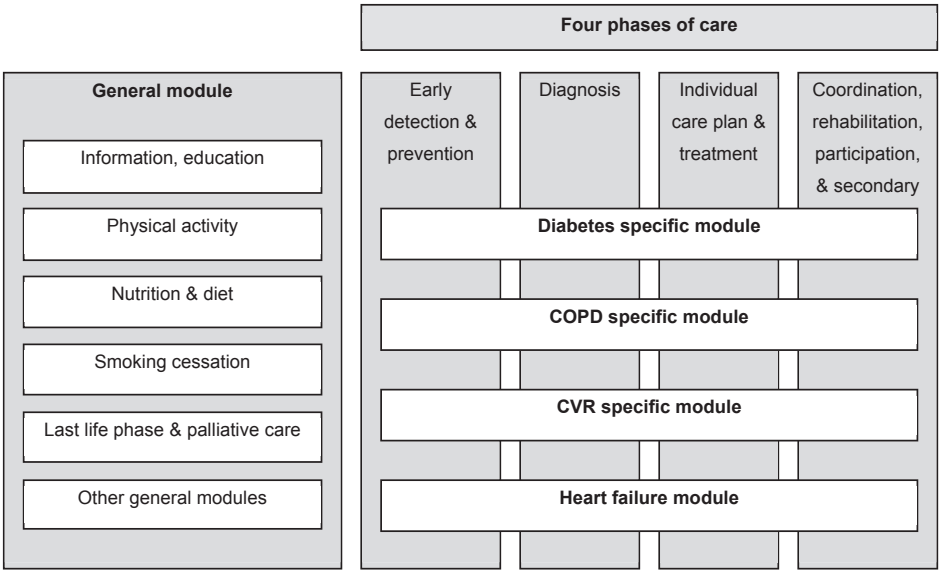
Figure 1 Schematic overview of integrated payment in the Netherlands



the opportunity to express the (dis-)satisfaction with the insurer's policy. The standard package is financed by a nominal premium paid directly by the insured and additional income-related premiums collected through a payroll tax. The latter premiums are distributed to health insurers based on a risk-equalization scheme to compensate for differences in the risk profiles of the insured population [18,100].

The standard insurance package is described in terms of health care functions (e.g. rehabilitation) and not providers (e.g. rehabilitation institute). In other words, the package is described in terms of 'what', 'when' and 'how' but not in terms of 'by whom' or 'where'. This is intended to stimulate managed care as insurance companies can purchase a particular type of care from any provider(s) able to deliver the care at good quality and a competing price (e.g. rehabilitation in physiotherapy practices) [101]. Health insurers are free to contract with providers that are registered according to the Individual Health Care Professionals Act (BIG). They negotiate with providers on price, volume, and quality of care. Insurers are also allowed to implement incentives to consumers to promote preferred providers, e.g. by offering premium rebates to patients who visit the preferred providers. This applies to both outpatient and inpatient care where health insurers can

Figure 2 Generic and disease-specific modules of the care standards



Source: ZonMw (2009), adapted by the authors

negotiate freely with hospitals regarding prices of a range of products called Diagnosis Treatment Combinations (DTCs). The DTCs were introduced in 2005 and are the Dutch version of the American Diagnostic Related Groups (DRGs) [76,102,103]. The percentage of hospital services offered to patients through DTCs for which the price is freely negotiated has increased from 10% to 34% in the last 4 years, denoting the effort of the Dutch authorities to promote regulated competition in the health care system [99].

3.2.2 Quality measures

To facilitate further regulated competition, the Dutch authorities have taken measures to improve information on quality of health care to support negotiations between health care purchasers and providers. These measures include the development of multidisciplinary evidence-based guidelines, process protocols and performance indicators. Patient involvement was substantial in the development of these measures in order to secure patient empowerment within the regulated competition [98]. Based on the guidelines and protocols, the Health Care Inspectorate (IGZ) develops indicators to measure health care quality in terms of structure, process and outcome performance [104,105] similar to the performance indicators developed by the Quality and Outcomes Framework (QOF) in the U.K. [16]. A representative selection of the performance indicators is provided in Appendix 1.

3.2.3 Financial incentives

There are also specific financial incentives to stimulate the development of appropriate structures and processes for the delivery of chronic care. Primary care providers can apply for additional reimbursement, categorized in three major modules, known as 'Innovation', 'Integrated Outpatient Care', and 'Modernizing and Innovation' [101]. These modules are granted to general practitioners who perform innovative interventions for chronic patients, promote multidisciplinary care teams in outpatient care, and set up DMPs. However, these incentives are segmented and do not ensure financial stability for sustained integration into the chronic care delivery system. Therefore, they will largely be replaced in 2010 by the new integrated payment system [106].

3.3 Barriers of the current policy

Although the current policies provide a framework to stimulate the integration of chronic care in the Netherlands, the success is still obscured by obstacles, mainly of a financial nature.

3.3.1 Segmentation of health insurance

Chronic care in the Netherlands is reimbursed through different health insurance schemes, depending on the type of care required: the Health Insurance Act (Zvw), the Exceptional Medical Expenses Act (AWBZ) and the Social Support Act (Wmo) [18,76,98,107]. For example, curative health care providers in primary care, outpatient and inpatient hospital care are financed by the Health Insurance Act; some types of home care are reimbursed by the Social Support Act (e.g. household help, transport support) and other types of home care by the Health Insurance Act (e.g. nursing care at home). Long term care (e.g. nursing home care, rehabilitation after cerebrovascular event) and social participation support are covered by the Exceptional Medical Expenses Act. As a result, from the patient's perspective, they have to knock on several different doors to get the care they need, and from the provider's perspective, multidisciplinary teamwork is hampered and an additional administrative burden is generated [108].

Furthermore, chronic care reimbursement is segmented not only across different health insurance schemes but also within a scheme. Some types of care for chronically ill patients that are recommended in the treatment guidelines are not included in the obligatory standard insurance package, only in the voluntary supplementary package. For example, preventive interventions such as pharmacological smoking cessation support or physical activity programs, are entirely excluded from the standard insurance package, while care provided by physiotherapists, and specialized nurses is only

partially included [108]. Consequently, the integration of care is dependent on whether or not a patient has voluntary supplementary insurance.

3.3.2 Suboptimal substitution between inpatient and outpatient care

In the current situation, health insurers have an (perverse) incentive to substitute outpatient care with inpatient care. The risk equalization scheme provides compensation retrospectively to health insurers for inpatient care only; thus, their financial risks related to inpatient care are reduced [105]. This differentiation in financial risks between the outpatient and inpatient care jeopardizes integration by reducing the attractiveness of the outpatient care market to health insurers. Considering the severity of this obstacle to integration, the Ministry of Health announced that the ex-post compensation will be gradually eliminated in the coming years [101].

3.3.3 Inadequate provider payment methods

In the Netherlands, the reimbursement of the general practitioners (GPs) is based on a mixed system consisting mainly of fee-for-service and capitation payment, while two thirds of all medical specialists are paid solely on (capped) fee-for-service basis [76]. However, it is argued that fee-for service is a fragmented payment method and is not suitable to integrated care [16].

Furthermore, the cooperation between care providers in outpatient care is not optimally reimbursed. The main contractor of a multidisciplinary team in outpatient care is eligible to receive further reimbursement through the additional reimbursement modules described in Section 2.3. In contrast, the sub-contracted providers, of the same multidisciplinary team, are not entitled to additional reimbursement and thus, the cooperation of providers is at stake. The main contractor might also receive a second payment for the same intervention because there is overlap between the interventions reimbursed on a consultation or capitation basis and the interventions that are listed on the modernizing and innovation modules [109].

3.3.4 Lack of information technology

The absence of an integrated Information Communication Technology (ICT) system is another obstacle to integrated chronic care in the Netherlands [31,110]. Although clinical information systems are partially implemented, the communication of information among them on a regional level is underdeveloped. With regard to outpatient care, there is no synchronization of information about what is delivered by the specialized providers for a chronic condition and what by general practitioners. Insufficient information flow exists also between the outpatient and inpatient care as well as between the different health insurance schemes (i.e. Zvw, Wmo, and AWBZ). Therefore, inefficiencies

such as unnecessary interventions and double payments for the same intervention are unavoidable and optimal cooperation of care providers is doubtful [101].

3.4 Introducing an integrated payment scheme

The Dutch Ministry of Health has introduced a new integrated payment system for chronic care in 2010 facing the most important (financial) barriers to integration of chronic care and realizing that sustainable funding of DMPs is still a challenge for integration in the Netherlands [31].

The Minister of Health has introduced a 'programmatic approach' towards chronic care in a series of letters to the Dutch parliament [101,106,111,112]. These letters describe the new payment system for integrated chronic care in detail. The new system is a prospective reimbursement system offering an 'all-inclusive' payment for people with chronic conditions to multidisciplinary teams of caregivers. Drugs, diagnostics and medical devices are not included in the payment yet, but this may change in the future. The new payment system is initially applied to four chronic conditions namely, diabetes, COPD, heart failure and management of cardiovascular risk factors (CVR). The relatively high prevalence of these chronic conditions and the feasibility of implementation were the main reasons for making this selection. The implementation started in January 2010 with diabetes and CVR and is planned to continue with the other two conditions later this year.

Under the new payment system, chronic care is coordinated by groups of providers (hereafter called care groups) that implement DMPs organized in integrated centers in primary care or in groups of cooperating general practices, paramedical care givers and/or hospitals [113]. Based on their structure, their legal form can be limited liability private company (BV), partnership, foundation, or limited partnership (CV) [114]. A schematic overview of the integrated payment system is presented in Fig. 1.

As Fig. 1 illustrates, health insurers purchase integrated care from care groups on the health care provider market by negotiating a fixed price per patient per year. This price is based on the expected case-mix of patients with a chronic disease. This price is called the chain-DTC because it combines costs of multiple professions mainly working in general or primary care and – to a limited extend – in specialized or hospital-based outpatient care. The care groups can deliver care themselves or sub-contract other providers on the health care provider submarket. The sub-contracting prices are subject to negotiation between the care groups and individual health care providers, such as general practices (see the bottom left branch in Fig. 1), primary care health centers specialists, nurses, and labs operating from hospitals (bottom middle branch), and/or individual providers (such as the dietician in Fig. 1). It should be noted that, up to now, the specialist has

been included in the chain-DTC only on a consultation basis. Specialized care provided in outpatient clinics and hospital wards is not part of the chain-DTC agreement. The chain-DTC primarily stimulates the cooperation between different providers of curative interventions in the primary care setting (e.g. GPs, GP practice assistants, physiotherapists, dieticians), but it does not intent to integrate the cure and the care sector, nor the hospital and primary care sector [113]. The price that is negotiated for a chain-DTC is not restricted by a minimum or maximum, although it is closely monitored by the National Health Care Authority (NZa), which is one of the two institutions through which the Ministry of Health (VWS) can control the chronic care market. The second institution is the Health Care Insurance Board (CVZ) which determines the insurance benefits included in the basic insurance package and therefore, indirectly influences the content of care negotiated between the care groups and the health insurers [101,114].

The negotiations on the content of care that is included in the chain-DTC are driven by the care as described in the 'care standards'. These care standards have been developed and authorized by caregiver organizations, patient associations, and public health authorities. Insurers are consulted during the development process of the standards. The care standards are mainly based on the existing medical guidelines, protocols, and performance indicators. The interventions described in the care standards are categorized into general and disease-specific modules as illustrated in Fig. 2. The general modules include interventions applicable to all chronic conditions, such as physical activity programs and smoking cessation support. The disease-specific modules are additions to the general modules, specifically targeted to each chronic condition. These disease-specific modules are sub-grouped along the four phases of care (see Fig. 2). The separation of interventions in these two types of modules prevents overlap of interventions for people with multi-morbidity since a person with two chronic conditions is offered the general modules only once. Performance indicators are attached to both general and specific modules to enable the monitoring of the delivered care. The recently introduced Minimum Data Set (MDS) by the Visible Care program (ZiZo) describes the set of performance indicators that must be collected anyway (see Appendix 1) [115]. As a result, the price of a chain-DTC may be driven by performance achieved and may differ according to the local agreements to stimulate improvement in these indicators.

Moreover, the integrated payment system aims to set a range of financial and non-financial incentives in place. First and foremost, the chain-DTC is a financial incentive for health care providers to improve the quality of care for patients with a chronic disease. By choosing from the different modules and adapting the modules to the specific patient needs, they can provide tailored care programs. Because self-management support is an important element of these programs, the integrated payment system contributes to the empowerment of patients with a chronic disease. Health insurers, by purchasing integrated care from care groups, can reduce financial risk, avoid double

payments, negotiate about prices within a trans-parent framework, and save costs due to the substitution of expensive inpatient care by less expensive outpatient care. Care groups, on the other hand, can generate more income by negotiating favorable prices with insurers, by increasing efficiency in care delivery, and by capturing a market share of chronic care that previously belonged to hospitals. Moreover, individual health care providers can reduce administrative costs because instead of negotiating with multiple health insurers they now negotiate with the care group only. Overall, care providers can improve their market position by providing attractive integrated care packages to people with chronic conditions and by scoring higher on performance indicators.

The new integrated payment system for chronic care in the Netherlands has some elements in common with payment systems for chronic care in other countries. In particular, it shares elements with payment for complete packages of chronic disease management care existing in the U.K. and Denmark. It also has similarities with the Medicare pay-for-performance initiatives in the U.S. where multidisciplinary practices are encouraged and financed based on performance indicators. Another similarity regards the German risk equalization formula, which since 2009 includes individual morbidity criteria [16]. However, none of these systems has promoted managed competition between insurers and among care groups by dividing the insurance purchase market into two submarkets, one in which insurers contract care groups and a second in which care groups contract providers.

3.5 Potential benefits

Although patient information on quality of care (i.e. quality consumer indices) is already provided by the National Institute for Public Health and Environment online (see www.kiesbeter.nl), the type of quality information published on this site depends on the availability of data and therefore differs per provider and health insurer [98]. In the new integrated payment system for chronic care, the care standards are positioned in the middle of the chronic care market and via the Minimum Data Set important quality information may be collected. As a result, quality may become more measurable and transparent not only for health insurers and care providers but also for patients. This may facilitate a better-informed choice of insurer and providers, which is expected to contribute to improvement of the quality of chronic care.

On the efficiency side, transparency of care budgeting through chain-DTCs enables health insurers to monitor their expenditures. Double payments are expected to decline because when a chain-DTC is assigned to a patient, then neither a 'hospital-outpatient' DTC nor a DTC for a related disease can be assigned to the same patient. For example, when a cardiovascular risk factors management (CVR) chain-DTC is assigned to a patient,

then a diabetes DTC or a heart failure DTC cannot be assigned to that patient any longer. There is no need to assign multiple DTCs, because the patient and provider can compile a package of care from the modules that is tailored to his or her specific needs.

Health care expenditures may further decrease by directly substituting care delivered in outpatient hospital clinics with primary care delivered through chain-DTCs. Hospital care may also be substituted indirectly when integrated care reduces the number of complications and hospital admissions. Moreover, efficient care substitution is anticipated to take place in the primary care setting too, because in chain-DTCs, physician assistants, specialist nurses and paramedical caregivers are given a more prominent role in performing tasks that have been traditionally performed by GPs. In the new payment system, GPs have agreed a pre-specified price with the insurers for the provision of chronic care and it is therefore, financially wise to delegate tasks to less expensive medical personnel. This might prevent situations similar to those found in Australia, Denmark and France where inadequate financial incentives to GPs for participating in multidisciplinary teams often hinder the delegation of tasks from GPs to other professionals [12].

Finally, the new payment system requires the development of ICT applications, such as integrated information systems and the minimum database, that are expected to improve the collaboration and coordination between care providers. This expectation is in line with the experience in England where a high level of computerization in general practices has improved the quality of care by providing individual care givers with the patient's entire treatment plan and facilitating case management [12].

3.6 Possible threats

The implementation of a major reform, such as the integrated payment scheme, is subject to potential threats to its success. It is important to identify and address these threats in order to avoid potential failures of the new payment system. The most important threats are presented below.

3.6.1 Limited choice of care

The patients' freedom of choice of insurer and provider has been extended over the last years through the promotion of managed competition [98]. This freedom might be jeopardized if a DMP contracts only a small number of providers, thus limiting the choice between different caregivers. It may also be jeopardized if a large DMP contracts with virtually all care providers in a region, thus limiting the patient's freedom not to participate in a DMP.

Limitations in the choice of care might also occur on the delivery side. Care providers might have limited freedom to provide treatments that are not agreed with the care

group. That could have a negative effect on the provision of tailored care to an individual patient with specific or exceptional needs.

3.6.2 Segmented integration

Although the recent policy developments in the Netherlands aim to integrate chronic care, there are risks for the contrary. First, Fabbricotti argued that DMPs might, in some cases, lead to fragmentation of care between contracted and non-contracted health care providers [116]. For instance, the continuity of care for people with chronic conditions depends on whether their current GP participates in the DMP contracted by their insurer. If that is not the case, the individual has to distinguish between general and disease-specific demand for care in order to seek care delivered by their current GP or by the DMP. Second, there might be fragmentation between different chain-DTCs or different DMPs. Considering that more than 50% of the Dutch population older than 55 has multi-morbidity [117], this risk of so called horizontal fragmentation is present, especially if DMPs address a single chronic disease. Third, there may be fragmentation between care included in the standard benefit package and care that is not. Some aspects of prevention, self-management, paramedical care and nursing care that are part of the care standard are not included in some DMPs because they are not reimbursed as part of the standard benefit package. Consequently, these forms of care get less priority than recommended in the care standards. This risk has been recognized by the Ministry of Health which has for example decided to include integrated smoking cessation support (pharmacotherapy in combination with counseling) into the standard package as of January 2010.

3.6.3 Market power misuse

Under the new payment system, care groups play a leading role in the chronic care market that might enable them to marginalize the role of non-contracted providers (see provider x in Fig. 1). Considering their weak bargaining power, non-contracted providers might face insufficient reimbursement from health insurers. The financial viability of non-contracted providers might further deteriorate if they are requested to help the high-risk population in case care groups would be able to exclude this population from the DMPs. Care groups might also use their market power to increase the price of chain-DTCs. For instance, health insurers might be prepared to pay a higher price in return for being the exclusive provider of a DMP that dominates a local market. If, however, exclusivity is prohibited, then DMPs might select patients from health insurers that pay the highest price.

Eventually, health insurers might attempt to overcome any excessive market power of care groups by developing their own DMPs similar to an U.S. HMO. This trend of vertical integration of health insurance and care provision has already increased during recent

years in the Netherlands [118,119]. Although, it is not yet clear what the results would be for the market position of the care groups and the individual providers, it could be expected that their market position would deteriorate.

3.6.4 Risk selection

Because the chronically ill are a substantial and increasing proportion of the population, health insurance companies may try to capture a larger share of this market. They can distinguish themselves from their competitors by contracting disease management programs on a large scale. However, if this distinction increases health insurers' popularity with the chronically ill too much, they risk disproportionally attracting patients in bad health conditions who have high health care costs. Hence, the development of disease management programs stresses the need to include a morbidity factor in the risk equalization formula for the allocation of funds to payers as it was introduced in 2009 in Germany [2]. The Dutch authorities have only now begun to develop this formula [16,100]. However, even when a morbidity factor is included in this formula, multi-morbidity is still not taken into account. Hence, insurers will still have an incentive for cream-skimming healthier persons and a disincentive to invest in chronic disease management for patients with multiple morbidities.

The risk of adverse selection is reinforced by the fact that health insurers can refuse to provide premium discounting to groups of people with chronic conditions that purchase health insurance collectively [100]. As long as some components of chronic care are not part of the standard health insurance package, there will be risk selection through the supplementary insurance package where health insurers can refuse coverage to applicants.

3.6.5 Organizational and management failure

Keeping in mind that care groups are involved in complex management tasks, potentially inadequate management competencies could hamper the successful implementation of integrated care. In addition, the negotiation-capability of care groups is the driving factor of price variation of chain-DTCs [120]. Therefore, the inabilities of a care group to negotiate adequately could distort the linkage between the price of a chain-DTC and the delivered quality (via the performance indicators).

Furthermore, care groups act both as purchasers (in the chronic care sub-market) and as suppliers (in the insurance market). This is likely to increase their administrative burden and to transfer bureaucracy from health insurers to care groups, which is a barrier to integration of care [121].

There are also concerns that delays in the implementation phase of DMPs will lead to reduction in the ambition to improve the content of care according to the chronic care model. Ambitions to develop new strategies to actively detect patients at risk, new

self-management strategies, new lifestyle interventions, and effective collaboration between providers might decline during a long implementation period. If these new interventions are not reimbursed or not developed because there is insufficient time, the chain-DTC will be nothing more than an alternative way of paying for the care that is already provided currently.

3.6.6 Costs and inefficiencies

Furthermore, double payments might still be present in the new payment scheme since partially contracted care providers are able to deliver services via both DMPs and individual practices at the same time. Consequently, these providers could be reimbursed twice for same care provision, once by the care group and once by the insurer.

There is also a risk in financing the registration of performance scores instead of the actual performance per se. As in pay-for-performance systems, care providers could 'game the system' and instead of providing better quality of care, they could focus on increasing the performance indicator scores [122]. This could be the case especially for providers with poor performance [28].

Finally, evidence suggests that disease management project leaders are not fully aware of the implementation costs of a DMP [123] and fail to anticipate them [23]. Consequently, the risk to underestimate these costs in the negotiated chain-DTC price might bring care groups in financial difficulties. Taking into account the considerable variation in the size of DMPs [114], this risk is higher for small DMPs that have relatively higher overhead and contract costs compared to large DMPs.

3.7 Necessary conditions for success

In the previous sections, we have outlined the urgency for success of the new payment system to put integrated chronic care in place and addressed the possible threats for failure. Based on these findings, we define a set of conditions necessary to fulfill the aim of the new payment system, which is listed as follows:

- To develop complete and adequate care standards with well-defined performance indicators necessary to secure high quality of care.
- To have written liabilities and responsibilities between health insurers, care groups, and individual providers that are linked to the standards of care.
- To include all components of integrated care provided by DMPs into the basic insurance package in order to avoid segmentation of care and risk selection of health insurers. For the entire programs to become part of the basic insurance package we need cost-effectiveness studies to support this.

- To make clear whether the total demand of a chronic patient is included in a disease specific chain-DTC or whether there are separate payment schemes for general and disease-specific care [124].
- To make the chronic care market attractive to health insurers in order to trigger them to act as purchasers of DMPs and develop the functionality of that market.
- To establish a transparent relationship between performance indicators and the price of the chain-DTC, to allow for differentiation in price based on the quality of care provided.
- To develop adequate information systems necessary to the success of disease management [17,125]. Adequate information is necessary to monitor the delivery of care, record performance indicators on an ongoing basis, facilitate the contracting between purchasers and providers and the coordination between providers. Information technology could also support the development of decision support systems available to health professionals and managers and provide patients and consumers with information necessary for rational purchasing in the chronic care market.
- To further stimulate the integration between primary care and specialist care provided in outpatient hospital clinics, the latter should become a more substantial part of the chain-DTC.
- To provide tailored care to patients with multiple chronic diseases by making a selection from general modules and disease-specific modules defined in the care standards. This calls for chain-DTCs for multiple morbidities, that can vary in price, depending on the selection of care modules. Desirably, that would be applicable for all chronic conditions and not only for close related diseases as diabetes, CVR, and heart failure. Hence, the segmentation between chronic diseases could be overcome.
- To introduce a (multi-)morbidity factor in the risk equalization formula might eliminate risk selection from health insurers.
- To have sufficient control and close monitoring of the chain-DTC prices and contracts between purchasers and providers by the Dutch authorities to secure adequate market functioning and avoid market failures.
- To evaluate the effects and costs of DMPs in order to provide information about the quality of care delivered and the costs attached to its provision to prevent inefficient care being included in the insurance package.

3.8 Call for evaluation

The last condition is crucial not only to the successful implementation of the new payment scheme but also to assess whether the adoption and promotion of DMPs as means to integration of care in the Netherlands is meaningful in the first place. There

is some evidence about the (cost-)effectiveness of DMPs. An example is the ELSID study in Germany, which concluded that DMPs improve the chronic care delivery to patients with diabetes according to the CCM [126]. Similarly, the INTERCOM trial in the Netherlands estimated that a community-based DMP for COPD patients can have an acceptable incremental cost-effectiveness ratio [127]. However this evidence is limited and inconclusive to support decision making on a large scale [2]. An economic evaluation of DMPs demonstrating cost-effectiveness could provide decision makers with hard evidence about the clinical and economic consequences of the new market model imposed on the chronic care market. Such an analysis could also support an optimal inclusion of treatments in the basic insurance package and it could enlighten health insurers about the short and long-term investment opportunities in chronic care and attract their attention to sustainably support DMPs. However, it should be recognized that the net return on investment in DMPs rarely occurs before 5 years of implementation [16]. An economic evaluation of DMPs would also inform care groups about the potential costs of the development and implementation of DMPs and the generated effects and therefore, could strengthen their negotiation with health insurers. Finally, people with chronic conditions would be ensured that the care they receive is of best qualitative and economic value.

The Dutch Organization for Health Research and Development (ZonMw) initiated and funded a large program for the development, implementation and evaluation of 22 DMPs in order to provide evidence about the (cost-)effectiveness of DMPs. These DMPs address a variety of chronic conditions such as diabetes, COPD, CVR, depression, anorexia, and psychosis and is the first nationwide attempt to evaluate DMPs in terms of effectiveness and cost-effectiveness. The economic evaluation is being conducted by the authors who are developing a standard framework for the evaluation and are using both program-specific outcomes as well as generic outcomes that enable comparison of results across the different DMPs [128]. The results will be available within three years.

Appendix 1 Representative selection of performance indicators included in the Minimum Data Set

Disease	Measurement category	Process indicator	Type of indicator
Diabetes II	HbA1c	% patients with a yearly HbA1c control	P
		% patients with HbA1c<7%	O
	BMI	% patients of which the BMI is measured in the last year	P
		%patients with BMI<25	O
	Ophthalmological examination	% patients that have a yearly ophthalmological examination	P
		%patients that received fundus control in the last 2 years	P
		% patients with retinopathy	O
	Feet examination	% patients that received a foot examination in the last 12 months	P
		% patients with foot impediment during the last examination	O
		% patients with measured Sims-score	P
	Blood pressure	% patients with a yearly blood pressure control	P
		% patients with SBP<140 mmHg	O
	Cholesterol	% patients with measured lipid profile	P
		% patients with total cholesterol <4,5 mmol/l	O
	Kidney function	% patients with a creatine measurement in the last 12 months	P
		% patients with creatine levels <60 and 30 mmol/l	O
		% patients with urine examination of albumine or albumine/creatine ratio in the last 12 months	P
		% patients with microalbuminurie	O
	Smoking	% patients with known smoking status	P
		% patients that are smokers	O
	Treatment	% patients with complete risk profile (hbac1, blood pressure, BMI, etc.)	P
		% newly diagnosed patients with a dietician consultation in the last year	P
		% patients with a dietician consultation in the last year	P
		Available expertise	S
		Availability of a specialized internist	S
	Quality of integrated care	% patients with an individual care plan	S
		% patients assigned to a central health care provider	S
	Other	% deaths in the last year (including age and death cause)	P
		% new patients with heart failure	O
		Total number of diabetic patients	S

Disease	Measurement category	Process indicator	Type of indicator
COPD	BMI	% patients of which the BMI is measured in the last year	P
	Treatment & monitoring	% patients with a spirometry test in the last year	P
		% patients with a known GOLD classification in the last year	P
		% patients with a known risk status (GOLD-score plus function status)	P
		% patients with a given inhalation instruction in the last year	P
		% patients with monitored CCQ or NCSI	P
		% patients vaccinated against influenza in the last year	P
		% patients with inhalation technique controlled in the last year	P
	Smoking	% patients with known smoking status	P
		% patients that are smokers	O
	Quality of integrated care	% patients with an individual care plan	S
		% patients assigned to a central health care provider	S
Cardiovascular risk	Blood pressure	% patients with a yearly blood pressure control	P
		% patients with SBP<140 mmHg	O
		% patients with SBP≥140 mmHg in the general practice population	S
	Cholesterol	% patients with measured LDL-cholesterol	P
		% patients with LDL-cholesterol,2,5 mmol/l	O
		% patients with total cholesterol ≥6,5 mmol/l in the general practice population	S
		% possible high-risk patients with a LDL measured in the last 5 years	P
		% patients with hypercholesterolemia in the general practice population	O
	BMI	% patients of which the BMI is measured in the last year	P
		%patients with BMI<25	O
		% high-risk patients with a measured BMI in the last year	P
	Smoking	% patients with known smoking status	P
		% patients that are smokers	O
		% patients male, older than 49 and smokers in the general practice population	S
		% patients female, older than 54 and smokers in the general practice population	S
	Alcohol	% high-risk patients with registered alcohol misuse behavior	P
	Quality of integrated care	% patients with an individual care plan	S
		% patients assigned to a central health care provider	S
	Other	% patients with glucose measured in the last 5 years	P

Disease	Measurement category	Process indicator	Type of indicator
		% high-risk patients in the age between 40-65	P
		% patients vaccinated against influenza in the last year	P
		% patients administered with anticoagulant medicine	P
		% patients with LDL-cholesterol ≥2,5 mmol/l administered with lipid lowering medicine	P+O
		% patients with basic risk-profile (smoking-status, SBP, LDL)	P

Source: Zichtbare Zorg (Visible Care), http://www.zichtbarezorg.nl/page/Eerstelijnszorg/Chronische-zorg; Note 1: P= Process, O= Outcome, S= Structure; Note 2: There are indicators common to multiple diseases; Note 3: BMI: Body Mass Index, Hba1c: Glycosylated Hemoglobin, SBP: Systolic Blood Pressure, GOLD: Global Initiative for Chronic Obstructive Lung Disease, CCQ: Clinical OPD Questionnaire, NCIS: Nijmegen Clinical Screening Instrument, LDL: Low-density Lipoprotein.

CHAPTER 4

Impact of financial agreements in European chronic care on health care expenditure

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Abstract

Various types of financial agreements have been implemented in Europe to reduce health care expenditure by stimulating integrated chronic care. This study used difference-in-differences (DID) models to estimate differences in health care expenditure trends before and after the introduction of a financial agreement between 9 intervention countries and 16 control countries. Intervention countries included countries with pay-for-coordination (PFC), pay-for-performance (PFP), and/or all inclusive agreements (bundled and global payment) for integrated chronic care. OECD and WHO data from 1996 to 2013 was used. The results from the main DID models showed that the annual growth of outpatient expenditure was decreased in countries with PFC (by 21.28 US$ per capita) and in countries with all-inclusive agreements (by 216.60 US$ per capita). The growth of hospital and administrative expenditure was decreased in countries with PFP by 64.50 US$ per capita and 5.74 US$ per capita, respectively. When modeling impact as a non-linear function of time during the total 4-year period after implementation, PFP decreased the growth of hospital and administrative expenditure and all-inclusive agreements reduced the growth of outpatient expenditure. Financial agreements are potentially powerful tools to stimulate integrated care and influence health care expenditure. A blended payment scheme that combines elements of PFC, PFP, and all-inclusive payments is likely to provide the strongest financial incentives to control health care expenditure.

4.1 Introduction

Chronic diseases account largely for the increase in health care expenditure due to their rising prevalence [8]. This impacts the overall economy because the direct and indirect costs of chronic diseases in health care are a sizeable share of a country's GDP [9]. As a result, chronic diseases constitute a great challenge to economies worldwide. This challenge is even greater when considering that chronic diseases may jeopardize economic growth through their impact on consumption, savings, labour productivity, labour supply, education, and life expectancy [2].

Improvements in chronic care management, coordination, and integration are seen as means to tackle increasing chronic care expenditure because of their potential to reduce costs while improving quality for people with complex health care needs [129,130]. Adequate financial flows and incentives are prerequisites to improving chronic care [28]. Thus, different financial agreements have been developed to provide appropriate incentives to the chronic care stakeholders. Similar to the US [43], adaptations of pay-for-coordination (PFC) (e.g. Austria, Denmark, France), pay-for-performance (PFP) (e.g. England, France), bundled payments (e.g. the Netherlands), and global payments (e.g. Germany) targeting different stakeholders (mainly health care providers and payers) have been introduced the last decade in Europe [16]. While these financial agreements and subsequent implemented policies have been described in the literature [16], their impact on health care expenditure has been poorly investigated.

The aim of this paper is to investigate the impact of different financial agreements of chronic care in Europe on national health care expenditures.

4.2 Methods

We used a difference-in-differences (DID) analysis to estimate differences, before and after (pre-post) introduction of a payment scheme, between countries that had financial agreements for improved chronic care (intervention countries) and countries that did not have any reforms in chronic care financing and/or payment (control countries). The DID analysis is used in econometrics to analyse panel data on country level to evaluate the impact of a certain event or policy [131]. Previous applications of DID in health care investigated the impact of medicine reimbursement [132] and hospital payment [133] reforms. We investigated the effect of financial policies that merely targeted the outpatient sector but, in some cases could involve hospital care as well.

We followed a three-step strategy to distinguish between intervention and control countries. We first searched in the literature to identify which European countries have taken large-scale initiatives towards providing financial incentives to stakeholders for

improvements in chronic care delivery. Second, we used published literature and WHO reports to ensure that control countries had not introduced financial agreements in chronic care [74,134]. Third, we conducted interviews in the intervention countries to gather more information about the financial agreements and to ensure that no other financial agreements on national level were implemented in the same year and country as the financial agreements of interest. A detailed description of these interviews and their results can be found elsewhere [42].

4.3 Data

We collected panel data from 1996 to 2013 for 25 European countries from the databases of WHO and OECD [135,136]. We used health care expenditure per capita corrected for purchasing power using current (August 2014) US$PPP. Our dataset consists of a) independent variables of interest, which are the chronic care financial agreements, b) outcome variables, and c) other covariates that control for cyclical and structural trends in each country.

4.3.1 Financial agreements

Traditional financial agreements, such as fee-for-service (FFS), capitation, and salary, are not designed to facilitate improvements in disease management, coordination of health care delivery and ultimately, integration of chronic care [16]. Therefore, countries with such agreements were considered as control countries in our analysis. We defined intervention countries as countries having one or more of the following financial agreements to support the improvement of chronic care delivery. These financial agreements could target individual care providers, a group of care providers in ambulatory care and/or secondary care, health insurers, and/or local authorities depending on the structure of a health care system. The first is pay-for-coordination (PFC), which involves payment to coordinate care provided by different types of providers for specified care services [70]. The second agreement is pay-for-performance (PFP), defined as the payment or financial incentive (e.g. a bonus) associated with achieving defined and measurable goals related to improvements in the process and outcomes of care for chronic diseases (e.g. patient experience, resource use, and other risk factors) [72]. The third category is the all-inclusive payment scheme. This category includes a) the bundled payment, which is a single payment for a group of services related to a specific condition that may involve multiple providers in multiple settings [73], and b) the global payment that is a single payment for the full range (i.e. not only disease-specific) of health care services needed by a specified group of people with chronic disease for a fixed period of time [43]. All-inclusive payments are commonly risk-adjusted.

4.3.2 Outcome variables

Similar to previous studies on payment reforms [132,133], we included a set of health care expenditure variables which we expected to be impacted by the payment reforms in chronic care. These outcome variables include total, outpatient, hospital, medication, and administrative expenditure per capita in health care. Outpatient expenditures include both primary care and outpatient hospital care. The administrative expenditures included expenditures for planning, management, regulation, collection of funds, and handling the claims of the delivery system.

4.3.3 Additional covariates

We added GDP per capita (expressed in August 2014 US$PPP) in our DID models to control for general economic trends per country. We also controlled for the total employment in the health care sector per thousand inhabitants to account for changes in the supply of health care per country.

4.4 Analytic modelling

We followed the work of Moreno-Serra and Wagstaff [133] and related literature [131,132] to specify our DID models. As suggested in the literature, our basic model was a simple DID model assuming that the trends in health care expenditure between countries and between intervention and control countries are parallel. This parallel trends (PT) model was estimated by taking the first differences (i.e. $X_t - X_{t-1}$ denoted as Δ) of all variables included in our analysis and is expressed as follows:

$$\Delta y_{it} = \beta \Delta X_{it} + \gamma \Delta PFC_{it} + \delta \Delta PFP_{it} + \theta \Delta ALL_{it} + \lambda_t + \Delta \varepsilon_{it} \quad (1)$$

where y_{it} is the outcome variable, x_{it} is a vector of additional covariates (i.e. GDP per capita, number of total employees in health care per 1000 inhabitants), and PFC_{it}, PFP_{it}, and ALL_{it} are dummy variables taking the value 1 when a country i at time t implements *PFC*, *PFP*, and *ALL* respectively. A period specific intercept is denoted as λ_t and ε_{it} is an idiosyncratic error term independent identically distributed over i and t.

The assumption of the basic model that the trends between the countries are parallel is relaxed in a random trends (RT) model, which is expressed as:

$$\Delta y_{it} = \beta \Delta X_{it} + \gamma \Delta PFC_{it} + \delta \Delta PFP_{it} + \theta \Delta ALL_{it} + \lambda_t + \kappa_i + \Delta \varepsilon_{it} \quad (2)$$

where κ_i is a country effect to allow for different trends among countries. The second assumption of the basic model about the parallel trends between intervention and control countries is relaxed in the differential trends (DT) model written as:

$$\Delta y_{it} = \beta \Delta X_{it} + \gamma \Delta PFC_{it} + \delta \Delta PFP_{it} + \theta \Delta ALL_{it} + \lambda_t + \psi_i + \Delta \varepsilon_{it} \quad (3)$$

where ψ_i is a group specific dummy indicating whether a country belongs to the intervention or control group of countries. In this model we control for common features that the intervention countries share and could have led to the adoption of financial agreements to improve chronic care.

The introduction of a financial agreement might not be fully in place during the implementation year or it might take some time before it influences health care expenditures. To test this assumption, we have also added time-lead variables in the three DID models for the three financial agreement variables. For example the dummy $PFC1_{it}$ takes the value 1 a year after the implementation of PFC in a country. Therefore, formula (1) becomes:

$$\begin{aligned} \Delta y_{it} = & \quad (4) \\ & \beta \Delta X_{it} + \gamma \Delta PFC_{it} + \delta \Delta PFP_{it} + \theta \Delta ALL_{it} + \pi \Delta PFC1_{it} + \tau \Delta PFP1_{it} + \\ & \varphi \Delta ALL1_{it} + \psi \Delta PFC2_{it} + \kappa \Delta PFP2_{it} + \zeta \Delta ALL2_{it} + \nu \Delta PFC3_{it} + \upsilon \Delta PFP3_{it} + \omega \Delta ALL3_{it} + \\ & \lambda_t + \Delta \varepsilon_{it} \end{aligned}$$

where $\Delta PFC(1,2,3)_{it}$, $\Delta PFP(1,2,3)_{it}$, $\Delta ALL(1,2,3)_{it}$ are the first differences of the financial agreement dummies in each of the 3 years after implementation of an agreement (e.g. $\Delta PFC1_{it} = PFC_{it+1} - PFC_t$). In other words, the dummy $\Delta PFC1_{it}$ takes the value 1 only the year after a PFC agreement was implemented. By adding these dummies a non-linear effect (i.e. an effect that is not equal in each year after the implementation) of a financial agreement can be captured. The linear combination of the differenced dummies (including the time-lead dummies) for each financial agreement was estimated to provide the cumulative impact of a financial agreement in the course of 4 years (implementation year plus 3 years afterwards). For example, considering PFC, the cumulative impact was estimated by summing up the estimates γ, π, ψ, ν and calculating the confidence intervals of the combined estimates.

The selection between the three models (i.e. PT, RT, or DT model), was based on their goodness-of-fit indicated by the Bayesian Information criterion (BIC). This criterion was used because it takes into consideration the number of predictors in a model and it is not influenced by the estimation of variance. We also looked at the joint significance of the country dummies in the RT model denoted as κ_i in Equation 2. If they were jointly insignificant the PT model was preferred to the RT model. Even if κ_i were jointly signifi-

cant but, uncorrelated with the financial agreement dummies then the PT model was still preferred. We tested this correlation using a generalised version of the Hausman test as demonstrated by Morenno-Serra and Wagstaff [133]. Further, if the group specific dummy ψ_i in Equation 3 was not significant then the PT model was preferred to the DT model.

A common problem in panel data is that the error term ε_{it} is potentially autocorrelated (i.e. correlated over *t* for a given *i*) and/or heteroscedastic. For these reason we used the Breusch-Pagan/Cook-Weisberg test to detect potential heteroscedasticity and the Langram-Multiplier test for autocorrelation. In case of heteroscedasticity we used robust estimation of variance and when autocorrelation was present we used cluster-robust estimation of variance [137]. We have also checked for multi-collinearity in the models using the variance inflation factor (VIF). The econometric analysis was performed in STATA 12.0 and was cross-checked using SAS 9.2.

4.5 Results

4.5.1 Financial agreements in chronic care

We identified 9 countries that had implemented financial agreements to support integrated chronic care, i.e. Austria, Denmark, France, Germany, the Netherlands, England, Portugal, Hungary, and Estonia. A short description of these financial agreements based on the literature and the interviews is presented below.

In Austria, the Health Reform Act of 2005 was implemented by the Ministry of Health to promote integration and coordination of care, improve efficiency, resource allocation and funding by pooling financial resources and promoting DMPs [32,33]. This reform created financial pools at state level by combining 1-2% of the budget of social health insurers with that of regional governments. These pooled funds were available for integrated care projects between primary and secondary care [74]. This was expected to overcome segmentation between the social health insurance scheme to fund outpatient care and the provincial health funds to fund inpatient care and to be economically beneficial for both schemes. The 2005 health reform act also promoted DMPs, funded by social health insurance, targeting general practitioners (GPs) and promoting their engagement in the coordination of integrated care efforts. On a national level, a DMP has only been implemented for diabetes, incorporating guidelines for cardiovascular risk assessment [75]. This was accompanied by a PFC payment scheme as physicians received an initial premium (€53) upon patient enrolment in DMP and a quarterly payment (€25) to supplement the traditional FFS. GPs qualified for providing DMPs if they participated in a basic training regarding care coordination, and attended refreshment courses. Additional courses on patient education were optional, for which physicians

would receive an additional remuneration. In Austria, there is no choice of insurer or competition among health insurance funds, as insurance is mandatory and contingent on place of residence or employer [32].

In Denmark, the Administrative Reform was initiated in 2007 to reallocate responsibilities between state, regions, and municipalities regarding decisions, financing and tasks [138]. This broad reform included the reallocation of responsibilities in the health care sector between five newly established Danish regions (reduced from originally 14 regions) and municipalities [23]. It was targeted at both regions and municipalities, promoting a PFC scheme on regional level. The idea was that a combination of regional coordinators, (non-) financial incentives and interdisciplinary care teams would improve continuity and coordination of chronic care services thereby increasing cost-effectiveness and quality of care provided [74]. Regions were provided with €70 million to improve chronic care by implementing comprehensive, well-evaluated programs, and municipalities were responsible for co-financing them [74]. Fifteen percent of each regional healthcare budget was allocated to integrated care activities, rewarding municipalities that successfully reduced the need for hospitalization through providing efficient preventive treatment and care [74,77,139]. Under the 2007 reform, municipalities were co-financing hospital care for their citizens and therefore had a financial incentive to promote preventive and integrated chronic care. There is no choice of insurer, and regions are responsible for organizing health services according to region specific demands and facilities [74].

In France, the Health Insurance Reform Act (2004) was an initiative targeting the primary care sector to promote the expanded use of DMPs. It was initiated as a negotiation between the social health insurance and the association of GPs. The aim of this program was to improve quality of care, patient monitoring, promote continuous medical education to communicate treatment guidelines to care providers, alleviate financial burden associated with unnecessary procedures, and strengthen the role of the GP [23]. The GPs got a supplement of €40 per patient enrolled in a DMP for care coordination, received self-education and training programs and experienced reduced waiting times in their practices. Further, CAPI (roughly translated to contracts for the improvement of individual practice) was launched as a voluntary pilot in 2009 and expanded in 2012 [84]. It was a PFP scheme in which GPs were rewarded financially for adequately registered patient records and for following evidence based guidelines. GPs could possibly get €6,000 annually (30% of their base salary), and 90% of them participated in the CAPI scheme in 2012.

In Germany, the Risk Structure Compensation Reform Act (2002) reaffirmed a global payment system targeting both primary and secondary care sectors. The wide implementation of DMPs in Germany began in 2003 when DMP registration became an additional risk adjuster [99]. This provided financial incentives to insurers to set-up as many DMPs

as possible and to contract as many physicians as possible because they could benefit up to €1,000 per patient by cutting down costs via the implementation of DMPs [80,82]. This risk-adjusted compensation aimed on one hand to avoid cream-skimming by the insurers at the expense of chronic ill patients and on the other hand to promote DMPs, which were believed to improve quality of chronic care [77,78]. Initially, DMPs existed for breast cancer, diabetes, coronary heart disease, asthma, and COPD and expanded to almost all chronic conditions [12]. Insurers who engaged chronically ill patients in DMPs were provided with additional remuneration for coordination costs (PFC) [77], in addition to the risk-adjusted compensation for health care coverage. The insurer could also reduce and/or waive co-payments to attract chronic patients to participate in DMPs [78]. Although this payment scheme targeted primarily health insurers with financial incentives and the remuneration of physicians did not change, care providers had an incentive to be contracted by insurers in order to increase their provided services. The Social Health Insurance-Competition Strengthening Act was implemented in 2007 to further strengthen and promote care integration [83]. It extended start-up financing for integrated care contracts until 2008, moved to include long-term care in integrated care contracts, and allowed non-medical healthcare professionals to contract with insurers [83]. In 2009, the risk adjustment scheme was updated to include morbidity indicators and thus, insurers receive higher remuneration for their chronically ill patients, regardless of DMP enrolment.

In the Netherlands, a bundled payment was piloted in 2007 with diabetes and expanded in 2010 to include COPD and cardiovascular disease management [47,89,90]. The aim of these payment reforms was to improve coordination between providers, promote the use of DMPs, strengthen adherence to medical guidelines, and increase quality of patient records [91]. Under the new payment scheme, chronic care is coordinated by groups of providers (called care groups) that implement DMPs organized in integrated centers in primary care or in groups of cooperating general practices, paramedical care givers and/or hospitals [47]. Insurers negotiated with care groups a predefined fee (bundled payment) that covered all care needed by a patient with a particular chronic disease for a year (excluding inpatient care, medication, medical devices, and diagnostics). Then care groups negotiate with and subcontract individual care providers for the care delivery [91]. Negotiations generate significant price variations between care groups for a particular group of patients i.e. different prices for different diabetes DMPs, serving to promote competition-induced quality improvements, on the basis of, but not limited to, performance measures, which are described in national care standards [47,93]. Insurers are free to choose whether they contract care groups based on the bundled payment system, or instead provide care groups only with an additional payment for the organization, coordination, and transparency of care, while continuing to reimburse individual providers on basis of a blended payment (i.e. capitation and

FFS). Patients are free to choose their GP and can change insurance company annually, choosing the most relevant, but least costly package to suit their medical needs [91].

In England, the Quality and Outcomes Framework (QOF) was introduced in 2004 [85]. The QOF offered PFP contracts to GPs, by which GPs were rewarded additionally based on 146 performance indicators within four domains; clinical standards, organizational standards, patient experience, and additional services [86-88]. This aimed to enhance the quality of primary care provided according to national guidelines, and its implementation was justified by the success of various quality-improvement initiatives that had been introduced since 1991 [86]. In 2006, adjustments were made to the system, altering minimum and maximum payment thresholds, dropping, modifying, and introducing new indicators [88]. In 2009/10, further adjustments were made, adding new indicators for heart failure, chronic kidney disease, depression, and diabetes, removing two indicators from the patient experience domain, and adjusting the point values of several indicators [87]. Initially, £1.8 billion was designated to reward GPs by a possible 25% salary increase, which was later increased to 30% [88]. Exception reports, through which GPs can decide to exclude patients from the calculation of certain irrelevant performance indicators, ensured a focus on relevant and appropriate targets [87,88]. Patients can use information, published by the NHS information center, to compare and choose a GP practice in which to enroll [88].

In Portugal, a 2006 reform updated the organization and funding mechanisms of primary care by introducing performance compensations (PFP), contracted annually and focused on vulnerable and high-risk patients [140]. Multidisciplinary teams (called USFs) rather than independent GPs were eligible, with targets relating to preventive care and chronic disease [141] . Their introduction aimed to improve care access, quality and continuity of care, satisfaction of care providers and patients and it was expected to lead to cost cuttings.

Moreover, in Hungary, the 1998 Act of Social Health Insurance Funds' Budget (effectively in place from 1999) reformed the payment system by introducing care coordination programs. In these programs, a coordinator (care organization, GPs, or hospital) was provided with financial incentives in the form of capitation fees (funded by social health insurance) to effectively manage the primary, secondary, and tertiary care for a specified population (initially up to 200,000 people) living in a specified area. This initiative was aborted in 2008 [74]. In 2009, performance and quality based indicators were used to employ a bonus payment system for family physicians. Initially €1.1 million were invested for GP bonuses, however this was increased nearly tenfold to €10.9 million by 2012 [142].

In Estonia, a PFP payment for ambulatory care was introduced in 2006, at which point there was 60% participation rate by GPs, which had increased to 80% by 2008 due to less stringent criteria for participation and increased financial incentives. The aim of this pay-

ment scheme was to increase the quality and effectiveness of preventive services and to improve the monitoring of chronic diseases. The PFP system targeted GP performance in three main areas; disease prevention, monitoring chronic conditions, and increased professional competency. GPs could get up to €255 per month on top of their usual per capita payment [143] for meeting the performance indicators [144].

Table 1 includes 25 European OECD countries classified as intervention countries if they employed PFC, PFP, and/or all-inclusive payments or control countries if they did not adopt any of these financial agreements. The year of payment scheme adoption refers to the year it became effective and not necessarily when the law was passed. When there was no information available about a country in the last years of our sample, we assumed that there was no change in the payment scheme considering that it would have been reported in the literature otherwise.

4.5.2 Impact of chronic payment reforms on health care expenditure

The results from the specification tests to determine the preferred model per outcome variable are presented in Table 2. The second column of this table shows that there is strong evidence for heteroscedasticity in almost all models, except for medication expenditure, indicating that robust estimations were necessary to be adopted in the analysis. In one model (outpatient expenditure) there was autocorrelation detected and therefore, cluster robust estimation was used. The BIC scores of each model are presented in the right half of the table. For the RT model the joint significances of the country dummies and the significance of the generalized Hausman test are also presented. The significance of the intervention group dummy is presented for the DT model. Based primarily on the BIC scores and cross-checked with the significance test, we selected the model (last column) that best fits each outcome variable (first column). The first panel of Table 2 presents the specification tests of the models without the time-lead variables and the second panel including them, as described in section 4.

The results from the different DID models are presented in Table 3 showing the impact of the introduction of a financial agreement on health care expenditure in the first year of implementation. The first column of the table shows the categories of health care expenditure per capita and their mean growth (for both control and intervention countries). None of the financial agreements did significantly reduce the growth in total health care expenditure. Countries with a PFC agreements had 21.28 US$ or 30% (= -21.28/70.86[1]) lower growth in outpatient expenditure. Moreover, countries which had introduced a PFP agreement had 64.50 US$ (87%) and 5.74 US$ (138%) less increase in hospital and administrative expenditure, respectively. The adoption of an all-inclusive agreement decreased the growth of outpatient expenditure by 216.60 US$ per capita

1. 70.86 is the mean increase in outpatient expenditure per capita per year in US$ in all countries

Table 1 Overview of financial agreements in chronic care per country by year

Year/Country	Classification	1997	1998	1999	2000	2001	2002	2003	2004	2005	2006	2007	2008	2009	2010	2011	2012	2013
Austria	Intervention									PFC	PFC	PFC	PFC	PFC	PFC	PFC	PFC	PFC
Belgium	Control																	
Czech Republic	Control																	
Denmark	Intervention											PFC	PFC	PFC	PFC	PFC	PFC	PFC
Estonia	Intervention										PFP	PFP	PFP	PFP	PFP	PFP	PFP	PFP
Finland	Control																	
France	Intervention									PFC	PFC	PFC	PFC	PFC, PFP	PFC, PFP	PFC, PFP	PFC, PFP	PFC, PFP
Germany	Intervention							ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC	ALL-G, PFC
Greece	Control																	
Hungary	Intervention			PFC	PFC	PFC	PFC	PFC	PFC	PFC	PFC	PFC	PFC	PFP	PFP	PFP	PFP	PFP
Iceland	Control																	
Ireland	Control																	
Italy	Control																	
Luxembourg	Control																	
Netherlands	Intervention														ALL-B	ALL-B	ALL-B	ALL-B
Norway	Control																	
Poland	Control																	
Portugal	Intervention										PFP	PFP	PFP	PFP	PFP	PFP	PFP	PFP
Slovak Republic	Control																	
Slovenia	Control																	
Spain	Control																	
Sweden	Control																	
Switzerland	Control																	
Turkey	Control																	
United Kingdom	Intervention								PFP	PFP	PFP	PFP	PFP	PFP	PFP	PFP	PFP	PFP

Control payments

FFS
salary
capitation

Intervention payments

ALL-G = all-inclusive: global budget
ALL-B = all-inclusive: bundled payment
PFC = pay-for-coordination
PFP = pay-for-performance

Table 2 Model specification tests for different outcomes

	Tests for selecting estimation of variance			Tests for selecting DID model						
		Estimation of variance		Parallel trends (PT)	Random trends (RT)			Differential trends (DT)		
	BP/CW* test	Langram** test				x_i	Hausman#		ψ_i	
	p-value	p-value		BIC	BIC	p-value	p-value	BIC	p-value	Preferred DID model
Health care expenditure										
				Panel A: main DID models						
Total	0.000	0.483	Robust	4612.98	4731.16	0.745		4678.77	0.847	PT
Hospital	0.012	0.429	Robust	3385.97	3485.51	0.677		3391.35	0.784	PT
Medication	0.279	0.439	Normal	3645.63	3767.77	0.002	0.878	3651.21	0.790	PT
Administrative	0.000	0.675	Robust	2226.36	2317.72	0.237		2231.88	0.883	PT
Outpatient	0.000	0.470	Robust	2278.64	2352.40	0.630		2283.03	0.455	PT
				Panel B: DID models with time-lead variables						
Total	0.000	0.319	Robust	4664.71	4782.70	0.767		4670.50	0.865	PT
Hospital	0.009	0.437	Robust	3434.95	3534.62	0.730		3440.42	0.905	PT
Medication	0.218	0.229	Normal	3692.56	3812.92	0.000	0.830	3697.45	0.538	PT
Administrative	0.000	0.719	Robust	2275.32	2366.55	0.279		2280.86	0.969	PT
Outpatient	0.000	0.033	Cluster	2324.60	2396.88	0.620		2328.89	0.494	PT

Note: in Breusch-Pagan/Cook-Weisberg for heteroscedasticity the H_0: constant variance; ** Langram test for serial correlation the H_0:no first order correlation; Note: Heteroscedasticity and serial correlation tested in basic model; κ_i : country dummies in Equation 2; ψ_i: group specific dummy in Equation 3 indicating intervention country or not; #If the p-value of the generalised Hausman test is smaller than 0.05 then the payment scheme dummies are correlated to the κ_i and therefore, the RT model should be chosen.

Table 3 Results from the main DID models

	PFC		PFP		All-inclusive				
	Estimate	Impact	Estimate	Impact	Estimate	Impact	N	R^2_{adj}	$\overline{VIF}$
Expenditure:									
Total (μ=116.98)	1.63	1%	-32.38	-28%	30.20	26%	335	0.58	1.81
Outpatient (μ= 70.86)	-21.28*	-30%	0.27	1%	-216.60***	-306%	173	0.33	3.63
Hospital (μ=62.75)	-17.68	-28%	-64.50***	-87%	-27.15	-43%	245	0.18	2.07
Medication (μ=15.43)	-10.92	-71%	3.88	36%	4.43	29%	304	0.26	1.83
Administrative (μ=4.16)	-0.40	-10%	-5.74***	-138%	2.33	56%	254	0.08	2.11

* p-value<0.05; **p-value<0.01; ***p-value<0.001; other covariates included in the model were: GDP, total employment in health care; the impact is calculated dividing the respective coefficient by the mean (denoted as μ) of the respective outcome variable

(306%). The model statistics in Table 3 point out that the preferred models explained 58% of the variance in total health care expenditure per capita, 33% in outpatient expenditure, 18% in hospital expenditure, 26% in medication expenditure and 8% in administrative expenditure in health care. There was no indication of strong multicollinearity since the mean VIF was close to 2 (>1.00) in almost alle models, but the VIF for each covariate was far below10.

Table 4 Results from the time-lead DID models

Expenditure:	Total		Outpatient		Hospital		Medication		Administrative	
	Estimate	Impact	Estimate	Impact	Estimate	Impact	Estimate	Impact	Estimate	Impact
PFC_t	4.43	4%	-22.74*	-32%	-20.61	-33%	-7.81	-51%	-0.29	-7%
PFP_t	-33.37	-29%	1.38	2%	-62.79***	-100%	4.35	28%	-6.15***	-148%
ALL_t	27.42	23%	-215.15***	-304%	-24.11	-38%	1.33	9%	2.23	54%
$PFC1_t$	-2.03	-2%	5.20	7%	9.24	15%	8.29***	54%	-2.87	-69%
$PFP1_t$	24.98	21%	2.21	3%	-40.10***	-64%	-5.61	-36%	-3.61***	-87%
$ALL1_t$	-70.53*	-60%	-19.55*	-28%	-12.51	-20%	-40.04***	-259%	0.45	11%
$PFC2_t$	38.90	33%	25.47	36%	-14.56	-23%	81.88	531%	0.25	6%
$PFP2_t$	-8.30	-7%	-17.44	-25%	-72.74***	-116%	21.55	140%	-5.10**	-123%
$ALL2_t$	30.30	26%	-83.37**	-118%	-37.80	-60%	-47.64	-309%	8.75***	210%
$PFC3_t$	-11.21	-10%	-27.70***	-39%	-18.36	-29%	11.86	77%	1.29	31%
$PFP3_t$	-43.56	-37%	-26.02	-37%	-37.67***	-60%	15.91	103%	-3.07**	-74%
$ALL3_t$	29.35	25%	40.17***	57%	37.71	60%	-13.86	-90%	-2.70	-65%
N	335		173		245		304		254	
R^2_{adj}	0.57		0.29		0.15		0.25		0.05	
$\overline{VIF}$	1.68		3.02		1.88		1.70		1.91	

* p-value<0.05; **p-value<0.01; ***p-value<0.001; other covariates included in the model were: GDP, total employment in health care; the impact is calculated dividing the respective coefficient by the mean of the respective outcome variable; ALL: all-inclusive

Table 5 Linear combined effect of each payment after 4 years of implementation

Expenditure	Payment	Combined estimate	Standard error	p-value	95% CI	
Total	PFC	30.10	60.19	0.624	-97.49	157.69
Total	PFP	-60.25	45.32	0.202	-156.33	35.83
Total	ALL	16.55	62.73	0.795	-116.43	149.53
Outpatient	PFC	-19.77	23.97	0.422	-70.58	31.04
Outpatient	PFP	-39.88	23.73	0.112	-90.20	10.44
Outpatient	ALL	-277.90	23.08	0.000	-326.83	-228.98
Hospital	PFC	-44.29	68.08	0.525	-188.61	100.03
Hospital	PFP	-213.30	14.61	0.000	-244.27	-182.33
Hospital	ALL	-36.71	66.34	0.588	-177.34	103.92
Medication	PFC	94.21	71.26	0.205	-56.84	245.26
Medication	PFP	36.20	18.97	0.074	-4.02	76.43
Medication	ALL	-100.21	71.51	0.180	-251.80	51.37
Administrative	PFC	-1.62	7.35	0.828	-17.20	13.96
Administrative	PFP	-17.92	1.90	0.000	-21.95	-13.90
Administrative	ALL	8.73	7.35	0.252	-6.85	24.31

ALL: all-inclusive

The results from the DID models including the time-lead variables for the three financial agreements are presented in Table 4. PFP financial agreements had a sustainable negative impact on (i.e. reduced) the growth of hospital and administrative expenditure, while all-inclusive financial agreements had a sustainable negative impact on the growth of outpatient expenditure. Moreover, the growth of medication expenditure was increased (by 8.29 US$ per capita) in countries with PFC agreements and decreased (by 40.04 US$ per capita) in countries with all-inclusive agreements in the year after implementation.

The combined effect of each financial agreement on the different expenditure categories during the four year after implementation is presented in Table 5. The cumulative effect of PFP during the four years after implementation on the growth of hospital and administrative expenditure was -213.30 and -17.92 US$ per capita, respectively. Moreover, the cumulative effect of all-inclusive payments on the growth of outpatient expenditure was a decrease of 277.90 US$ per capita.

4.6 Discussion

This paper has identified European countries that have implemented financial agreements to facilitated integrated chronic care and provided tangible evidence of their impact on health care expenditure.

The results from the main DID models showed that at the year of implementation, PFC decreased the growth of outpatient expenditure while PFP decreased the growth of hospital and administrative expenditure. Further, countries with all-inclusive financial agreements had a decreased growth of outpatient expenditure. Considering the cumulative impact of the financial agreements on all expenditure categories in the four years following their implementation, PFP decreased hospital and administrative expenditure while all-inclusive agreements decreased outpatient expenditure.

It is noteworthy that the three types of financial agreements had an immediate impact on outpatient, hospital or administrative expenditure and in many cases this impact continued in the following three years after implementation. In contrast, the impact of financial agreements on total and medication expenditure was delayed. However, in the case of all-inclusive agreements the impact in the first three years was not in the same direction as the impact in the fourth year after implementation. This indicates that it takes more than a year for the effects to become fully apparent and that the effects may decrease over time. This may also raise questions about actions taken by insurers or health care providers to compensate for any intended cost reduction.

Moreover, PFC appeared to substantially decrease outpatient expenditure (by 30%) at the year of implementation. This is in accordance with the literature that found care coordination programs cost-saving [145]. In contrast, PFC increased the medication expenditure a year after the implementation year. This might be explained by optimization of drug treatment and improved adherence due to early diagnosis and monitoring of chronic patients achieved by better coordination of care providers. Furthermore, PFC agreements were in many cases combined with all-inclusive or PFP agreements. These combinations might be interesting to take into consideration when reforms in chronic care are designed because one financial agreement might compensate for the potential adverse incentives of another financial agreement.

Furthermore, PFP appears to have the strongest ability to tackle the growth of hospital. This might be attributed to direct financial incentives that PFP provides to providers to improve quality of care delivered, enhance efficiency, and encourage collaboration between primary and secondary care [70]. Walker et al. [94] demonstrated that the PFP agreements implemented in the UK was cost-effective in terms of cost per quality adjusted life years. However, a systematic literature review of PFP agreements in chronic care only showed small positive effects on quality of care but, there was no evidence available for the effects on health care costs [146]. Previous experiences with such finan-

cial agreements in the US might raise some concerns about the full reliance on PFP and therefore, it is suggested to be implemented as a valuable component of a more global payment model [43,147] such as bundled payments [72].

The all-inclusive financial agreements appear to decrease the growth of outpatient expenditure substantially. It should be explicitly mentioned that the bundled payment in the Netherlands targeted health care providers, while the global payment in Germany primarily provided financial incentives to health insurers. Although, there were concerns about the implementation of global payment in Germany with regards to supply induced demand [82], distorted competition between health insurers [80], and its volatile implementation [81], a recent empirical study found cost-savings after its implementation [148]. Concerning the recent implementation of bundled payments in the Netherlands, there is little evidence about its impact on health care expenditure and a debate about moving to a global payment system with shared-saving incentives is in process [93] following recent examples from the US [149].

This study investigated the impact of financial agreements on health care expenditure only three years after their implementation. This time-window was selected based on the data availability after the last implemented agreement of interest (i.e. bundled payment in the Netherlands was introduced in 2010 and the dataset extends up to 2013). However, the effects of integrated care in terms of reduced hospitalizations and complications are expected to increase in the longer term. Adequate financial agreements are necessary to provide the right incentives to achieve integration of chronic care [74] and therefore, the cost-reducing potential of integrated chronic care will not spread widely in the health care system without substantial change in financial arrangements [129].

The strengths of this study include the innovative character of analysing empirically the impact of financial agreements for integrated chronic care on health care expenditure, the adoption of DID models that are suitable for evaluating policies on country level, and the combination of qualitative and quantitative research methods that give a comprehensive understanding of the financial agreements and their impact. Nevertheless, this study has also limitations. First, we could not control for the non-financial policies but we assumed that these policies would be equally probable to have been implemented in the intervention and control countries. This assumption is supported by the fact that the DT model, which distinguished the health care expenditure trends between intervention and control countries, did not have a better fit. Second, it was not possible to distinguish between the impact of the bundled payment system and the global payment system because there was only one country that had implemented each one of them and in the case of the bundled payment, the observations were limited to only the last three years of the dataset. Third, we did not assess the impact of financial agreements on other outcomes, such as health outcomes, continuation of care, and implementation of guidelines, due to the lack of data. Fourth, we did not include an age-

structure variable in our models to control for healthcare demand because the missing observations in the variable "percentage of population older than 65 years" reduced the sample size used in the regressions too much. However, we run the models including this variable and the results remained the same.

Future research may investigate the impact of financial agreements on a wider range of outcomes in the longer term using a larger dataset with more observations after implementation of a payment scheme.

4.7 Conclusions

Financial agreements are potentially powerful tools to stimulate the delivery of integrated care and influence health care expenditure. PFP and all-inclusive financial agreements have the potential to reduce substantially the growth of health care expenditure. It also appears that PFC has the potential to reduce outpatient health care expenditure. Therefore, a blended payment scheme that combines elements of PFC, PFP, and all-inclusive payments is likely to provide the strongest financial incentives to control health care expenditure. Such a payment could follow examples from the US where payments include a yearly risk-adjusted tariff for the costs of chronic patients in primary and secondary care (as in global payments), cover the costs of coordination between care providers (as in PFC), and depend on performance indicators (as in PFP) following the footsteps of initiatives in the U.S [150].

PART B

Cost variation in disease management programs

CHAPTER 5

Exploring the variability of patient costs in disease management programs: a hierarchical modelling approach

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Abstract

Disease management programs include a wide variation of patients with different chronic diseases and different health care utilization. The aim of this article was to identify factors on patient-level and organizational-level that explain the variability in costs of patients with different chronic diseases enrolled in a DMP by employing a rigorous analytical model. A generalized linear mixed model (GLMM) was specified to perform a multi-level analysis of cross-sectional hierarchical data from 16 DMPs in the Netherlands. Multiple imputation, sub-group analysis per disease and analysis from both the health care and the societal perspectives were also performed. Our model showed that age, the presence of cardiovascular disease, multi-morbidity and payments on top of the payment for the usual care had positive relation with costs, while better quality of life was associated with lower health care costs. In the COPD sample, physical activity and employment were associated with health care costs. Our study showed that there is great variability in health care costs among patients included in DMPs and identified patient and organizational explanatory factors. The findings are relevant to the design of future DMPs and their payment schemes.

5.1 Introduction

Many governments around the world have promoted the implementation of disease management programs (DMPs) as a means of improving the effectiveness and efficiency of health care for the growing number of patients with a chronic disease [23,47]. Schrijvers (2009) defines these programs as 'a group of coherent interventions designed to prevent or manage one or more chronic conditions using a systematic, multidisciplinary approach and potentially employing multiple treatment modalities' [24].

Many European countries have implemented payment schemes to support the development and implementation of DMPs [151]. In the Netherlands, a bundled payment scheme was introduced in 2010. Under this scheme, health care insurers purchase chronic care from DMP providers based on a predefined price for an expected case-mix of chronic patients. This price is negotiable between the two parties, it is paid on a yearly basis per patient included in a DMP, it is not risk-adjusted and it includes mainly primary care and coordination costs [47].

Apparently, apart from the intervention provided by a DMP, the case-mix of patients included in a DMP determines the financial viability of DMP providers. This is because the negotiated price could be lower than the actual costs, if the case-mix of patients is not correctly anticipated. In other words, the variation of participants in DMPs could cause large heterogeneity in health care costs of DMPs. Therefore, the relation patient case-mix with health care costs should be taken into account from DMP providers when negotiating a fixed price per patient. Although there are different strategies to identify the risk profiles of patients for enrolment in DMPs, using a combination of both clinical and economic criteria, applications in the literature can be barely found [152]. From an economic perspective, this is amplified by the weak statistical analysis of cost data in previous studies of DMPs. Most studies have only studied the impact of patient-level characteristics on costs [55,61] or they have not considered the usually skewed health care cost data [63,153].

The aim of this article is to identify factors on patient-level and organizational-level that explain the variability in costs of patients with different chronic diseases enrolled in a DMP by employing a rigorous analytical model. We focus on Dutch patients with an increased cardiovascular risk profile (CVR), chronic obstructive pulmonary disease (COPD) or diabetes mellitus (DM).

5.2 Methods

5.2.1 Theoretical model of factors affecting health care costs

The selection of relevant covariates included in the analysis was based on the model developed by Andersen and Newman (2005), which was further adjusted for the purpose of our analysis [154]. The model assumes that the level of health care utilization per person depends on (1) predisposing factors including demographic, social-structural and attitudinal-belief factors, (2) enabling conditions that make health service resources available to the individual and (3) (perceived) illness level.

As predisposing factors, we included age, gender, educational level, employment and marital status. We extended the predisposing factors by adding behavioural factors including level of physical activity, smoking status and self-efficacy [155]. Based on previous evidence, we hypothesized that older patients [156,157], females [158,159], current and former smokers [160], single persons [161,162] and individuals with lower educational level [163] have higher health care costs. In contrast, more physical activity [164], greater self-efficacy [165,166] and being employed [167] are negatively related to health care costs.

We also adjusted the enabling factors of Andersen and Newman's framework to make them suitable for cost analysis in DMP's keeping their original principles. Consequently, we defined enabling factors as elements that enable chronic patients to receive adequate and high quality, multidisciplinary and integrated chronic care. These elements include the level of disease management and the presence of supplementary funding for providing integrated care. Level of disease management was measured by the Patient Assessment of Chronic Illness Care (PACIC) [168]. Additional funding is defined as payments that are received on top of the payment for the usual care to stimulate integration of care. There is no clear evidence about the impact of these aspects on health care costs [59]. However, we hypothesized that the enabling factors have a positive relation with health care costs, at least in the short term, as they enable the utilization of more comprehensive chronic care that was not provided previously.

As illness level factors we chose quality of life as measured by the EQ-5D using the Dutch utility values [169], multi-morbidity as measured by the updated Charlson comorbidity index [170], and the presence of a diagnosed cardiovascular disease (CVD). We hypothesized that quality of life has a negative relation with health care costs [171] while multi-morbidity and CVD have a positive relation with health care costs (Wolff et al., 2002). We also included the target population of a DMP and expected that patients in COPD-DMPs have higher health care costs than patients in CVR- or DM-DMPs. The definition, operationalization and range of scores of the covariates used in the analysis are presented in Table 1.

Table 1 Covariates used in the analysis: definition and measurement

Determinants	Patient-level	Organization-level	Measurement and operationalization	H_1*
Predisposing	Age		Age of the respondent in years at the time of questionnaire completion	+
	Smoking		A series of three dummy variables for smoking (never smoker, current smoker and ex-smoker - reference group is never smoker)	+
	Gender		0 male; 1 female	+
	Educational level		0 high education; 1 low education (defined as no or only primary education)	+
	Physical activity		Number of days per week physical active for more than 30 minutes	–
	Employment		0 not employed; 1 employed	–
	Marital status		0 not single; 1 single (including never married, divorced, widow/er)	+
	Self-efficacy		Component of the Self-Management Ability Scale with 0 being worst and 100 being best score [155]	–
Enabling	DM level (patient perspective)		PACIC measurement scale with 1 being worst and 5 being best score [168]	+
		Additional financing	0 no additional payment for integrated care and 1 additional payment or bundled payment [47]	+
Illness level	Quality of Life		EQ-5D utility scale, -0.59 (lowest) to 1 (highest) [169]	–
	Multi-morbidity		The Charlson comorbidity index, 0 (lowest) to 42 (highest) [170]	+
	Cardiovascular disease		0 without existence of diagnosed cardiovascular disease; 1 with existence of diagnosed cardiovascular disease	+
	CVR/DM-DMP		0 not included in a CVR- or DM-DMP (thus included in a COPD-DMP); 1 included in a CVR- or DM-DMP	–

* Research hypothesis (H_1): relation of covariates with health care costs is (+) positive or (–) negative.
DM: Disease Management, PACIC: Patient Assessment of Chronic Illness Care, EQ-5D: Euro-QoL 5 dimensions

5.2.2 Data and settings

The data was collected as part of a large evaluation study of 22 DMPs in the Netherlands [128]. It was a cross-sectional study of patients who have enrolled in DMPs. On the patient-level, a questionnaire was distributed to individuals recently enrolled in 9 CVR-, 4 COPD- and 3 DM-DMPs. The remaining DMPs addressed other diseases not included in this study. The CVR-DMPs included patients who were at risk for developing CVD and patients who had already been diagnosed with CVD. The questionnaire asked about health care resource utilization (including healthcare provider contacts, hospitalization and medication) in the past 3 months, quality of life, quality of care, self-efficacy, physical activity and socio-demographic characteristics. In addition, questions about absence

from paid employment or reduced productivity due to illness while at work, and travelling expenses of patients to the healthcare providers were asked. Information about the presence of additional funding to cover the specific elements of integrated care was collected separately during interviews with the head managers (project leaders) of the DMPs.

In our analysis, the health care costs from the health care perspective included the costs of (a) healthcare provider contacts including general practitioner (GP), nurse, assistant GP, specialist, physiotherapist, dietist, podotherapist, speech-therapist, ergotherapist, (b) hospitalization including intensive care units and (c) medication for CVR, COPD and DM. The costs of travelling to care providers born by patients and the costs of productivity loss due to illness estimated using the friction method [172] were added to estimate the costs from the societal perspective. All costs were inflated to 2012 values.

The 16 DMPs were spread across different regions of the Netherlands, but they were all managed by care groups in primary care or in outpatient clinics of hospitals [128]. Although the services included in the integrated care package differed between the DMPs, most programs included collaboration between different disciplines of health care professionals and a redesign of the care-giving process to improve chronic care management. They provided mostly interventions such as self-management education and training directed at life-style improvement, support with implementation of guidelines and protocols, integration of information systems, training for health care providers, case management and reallocation of tasks between care providers [128].

5.2.3 Statistical analysis

Generalized linear mixed models (GLMM) were used to perform a multi-level nonparametric analysis. This analysis was performed to facilitate (a) covariates on patient and organizational-level, (b) correlated health care costs of patients clustered in a DMP and (c) traditionally right-skewed health care costs data. During the last decade, GLMMs have been increasingly used in health care literature [173-177]. GLMMs were also selected because they overcome limitations of OLS and two-stage models by avoiding with ad-hoc adjustments for skewed data and accommodating zero observations for costs simultaneously [157,178-180,180-183]. The GLMMs were specified using unstructured covariance matrix, lognormal distribution and identical link function. A detailed description of the model selection is provided in Appendix.

Three models were specified in the main analysis using the complete case sample (n = 1906). Model 1 consisted of the random intercepts without any covariates. In Model 2 all covariates (as presented in Table 1) and the random intercepts were included to investigate their effects on health care costs simultaneously. Model 3 was specified starting from an empty model (only random intercepts) and then adding covariates one-after-the-other to get the model that best fits to the data. The inclusion of a covari-

ate was based on the likelihood ratio (or deviance) test that is suitable to compare the goodness-of-fit of models with nested covariate structure and same covariance matrix, distribution function and link function [184]. We first included the covariates of Model 2 that were not significantly related to health care costs. This was done to examine which of these covariates had a significant effect on health care costs and which had not, before adjusting for other covariates that were highly significant.

Furthermore, we performed a sub-group analysis by applying Model 2 to the three different disease populations included in the analysis, namely Model2-CVR, Model2-COPD, Model2-DM. Model 2 is selected for the sub-group analysis by disease because we were interested in investigating the effects of all covariates on health care costs in the three patient groups. Similarly, we used Model 2 to investigate the differences in the effects of all covariates between the societal perspective and the health care perspective. The model for costs from the societal perspective was named Model2-SP.

Regarding missing data, we performed a descriptive analysis of the cost data for the covariates that had missing values to explore the potential bias, if the incomplete cases (n = 952) were excluded from the analysis. A multiple imputation using the Markov Chain Monte Carlo (MCMC) method was used to impute the missing observations [185]. The imputation was performed separately for each type of disease and DMP and generated 10 complete data sets. Then we fitted model 2 and 3 in our analysis to the 10 imputed data sets named Model 2-MI and Model 3-MI, respectively. All analyses were performed in SAS 9.2.

5.3 Results

5.3.1 Sample characteristics

The sample characteristics by disease are presented in Table 2. The COPD sample includes proportionally more smokers and ex-smokers, patients with low educational level, unemployed patients and single patients than the other two samples. COPD patients are older, have higher Charlson co-morbidity scores, lower quality of life and self-efficacy scores, and higher costs compared to the CVR and DM samples. Compared to the DM sample, the CVR sample contains more patients with a cardiovascular disease, but the Charlson co-morbidity scores and the costs are lower. On the organizational-level, all DM-DMPs receive some form of additional funding for integrated care.

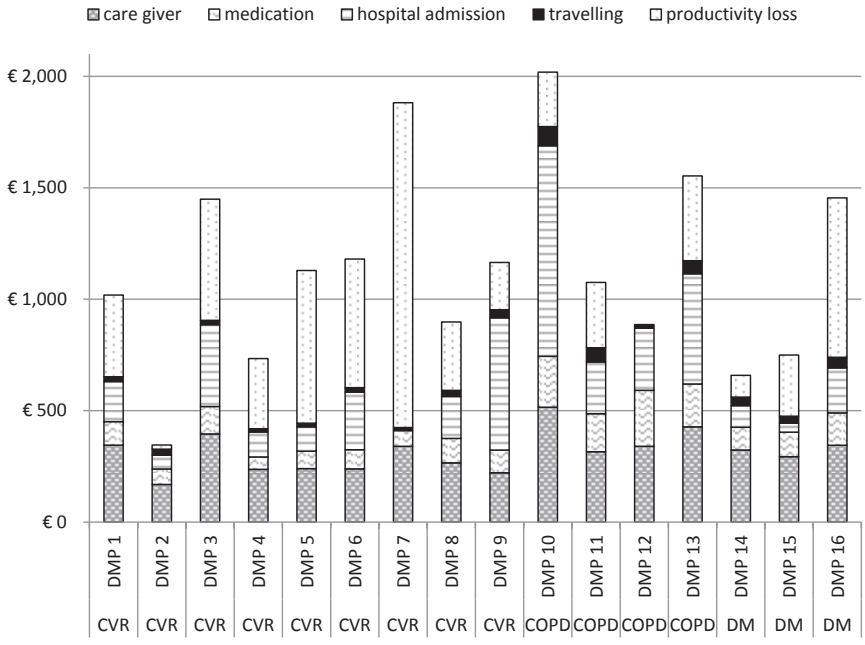
Figure 1 presents the health care cost categories included in the analysis by DMP. There is large variability in the total health care costs of patients and the break-down of these costs into different categories.

Table 2 Sample characteristics by disease

	CVR	COPD	DM	Total
	(n_p=1538,rr=46%, n_{DMP}=9)	(n_p=909, rr=60%, n_{DMP}=4)	(n_p=411, rr=59%, n_{DMP}=3)	(n_p=2858, rr=52%, n_{DMP}=16)
Age**	64.0 (10.2)	66.1 (10.6)	64.6 (10.2)	64.7 (10.4)
Females	48 %	47%	44%	47%
Smokers**	20 %	39 %	21 %	26 %
Ex-smokers	46%	48 %	43 %	46 %
CVD**	24 %	13 %	10%	19 %
Charlson index**	1.6 (1.6)	2.6 (1.7)	2.5 (1.7)	2.1 (1.7)
Low educational level**	39 %	49 %	25 %	40 %
Employment**	42 %	33 %	40 %	39 %
Single*	26 %	33 %	30 %	29 %
Physical activity	4.9 (2.1)	4.7 (2.2)	4.7 (2.1)	4.8 (2.2)
EQ-5D**	0.81 (0.20)	0.77 (0.22)	0.81 (0.20)	0.80 (0.21)
PACIC**	2.8 (0.9)	2.9 (0.9)	3.2 (0.8)	2.9 (0.9)
Self-efficacy*	68.5 (16.3)	67.2 (17.8)	69.7 (17.0)	68.3 (16.9)
Additional financing	33%	75%	100%	56%
Total costs HC	585 (2485)	1008 (2874)	540 (851)	713 (2468)
Total costs SP	889 (3003)	1268 (3210)	766 (1635)	992 (2924)

* p< 0.05 **p< 0.01; The table presents the mean (SD) unless otherwise indicated; CVD: cardiovascular disease, SES: socio-economic status, PACIC: Patient Assessment of Chronic Illness Care, EQ-5D: Euro-QoL 5 dimensions, HCP: health care perspective, SP: societal perspective, rr: response rate

Figure 1 Break-down of 3-month health care and total costs by DMP



5.3.2 Descriptive statistics

The results from the descriptive analysis are summarized in Table 3 and show that males, smokers, patients with a cardiovascular disease, people with low educational level, unemployed and single had significantly higher costs than their counter groups from the health care and societal perspective. In addition, age (only from the health care perspective), multi-morbidity had a significant positive association with health care utilization and total costs, while physical active days, EQ-5D and self-efficacy were negatively correlated with costs. PACIC scores were not significantly correlated with health care costs.

Table 3 Health care costs by sample characteristics and perspective

	Health care perspective mean (sd)	Societal perspective mean (sd)		Health care perspective Spearman correlation with costs	Societal perspective Spearman correlation with costs
Gender #					
female	661 (1688)	870 (2092)	Age	0.11**	0.16
male	746 (2960)	1099 (3485)			
Smoking**			Physical activity	-0.08**	-0.09**
never	591 (2016)	847 (2561)			
quit	704 (1858)	945 (2337)	EQ-5D	-0.41**	-0.38**
yes	890 (3680)	1287 (4117)			
CVD**			PACIC	-0.01	0.02
yes	1286 (4318)	1664 (4726)			
no	581 (1761)	837 (2288)	Self-efficacy	-0.14**	-0.14**
Low educ. level**					
yes	779 (2177)	1032 (2687)	Charlson index	0.47**	0.44**
no	691 (2748)	993 (3171)			
Employment ##					
yes	592 (1888)	1264 (3102)			
no	800 (2837)	847 (2887)			
Single*					
yes	889 (3760)	1169 (4021)			
no	647 (1684)	928 (2344)			

* p<0.05 ** p< 0.01 # p<0.05 from health care perspective and p<0.01 from SP ## p<0.01 only from health care perspective

5.3.3 Results from the GLMM using complete cases

5.3.3.1 Main GLMM results.

The results from the analysis of the complete cases (n_p=1,906, n_{DMP}=16) are presented in Table 4. Model 1, which is the model without covariates, had an intraclass correla-

Table 4 Results from the complete case analysis

	Model 1		Model 2		Model 3		Model 2-SP	
Fixed effects	exp(β)	p	exp(β)	p	exp(β)	p	exp(β)	p
Intercept	290.67	<.001	571.58	<.001	355.49	<.001	676.61	<.001
Physical activity			1.01	0.640			1.01	0.501
Employment			0.94	0.364	0.93	0.312	1.29	0.001
Gender			1.06	0.511	1.04	0.597	0.98	0.798
Age			1.01	0.024	1.01	0.017	1.00	0.742
Smoker			0.95	0.496			0.99	0.926
ex-Smoker			1.04	0.550			1.04	0.646
CVD			1.43	<.001	1.43	<.001	1.48	<.001
Charlson index			1.28	<.001	1.28	<.001	1.29	<.001
Low educational level			0.94	0.156			0.93	0.285
Single			0.98	0.777			1.05	0.501
EQ-5D			0.26	<.001	0.27	<.001	0.26	<.001
PACIC			1.02	0.356			1.06	0.110
Self-efficacy			1.00	0.651	1.00	0.919	1.00	0.211
Additional financing			1.17	0.046	1.18	0.063	1.16	0.046
CVR/DM-DMP			0.69	0.001	0.73	0.008	0.72	<.001
Random effects								
σ_e^2 (residual)	1.7166		1.4674		1.4705		1.8269	
σ_{u0}^2 (intercept)	0.1309		0.005081		0.007998		0.001716	
ICC	7%		0.3%		1%		0%	
R_1^2			20%		20%		16%	
R_2^2			85%		83%		82%	
AIC	6479.71		6181.76		6173.53		6597.2	

Note: (n_p=1,906, n_{DMP}=16); ICC = $\sigma_{u0}^2 / \sigma_{u0}^2 + \sigma_{e0}^2$

tion coefficient (ICC) of 7%, which means that 7% of the total variation in health care costs was due to variation in the mean costs of the 16 DMPs. In Model 2, which is the saturated model, the ICC had dropped to 0.3%, indicating that much of the variation in health care costs had been explained. The R_1^2 of this model was 20%, indicating that 20% of the variation in health care costs between individuals was explained. The R_2^2 was 85%, indicating that 85% of the variation in health care costs between the DMPs was explained. Age, the presence of a CVD, the Charlson co-morbidity score, the EQ-5D utility, additional funding for integrated care and CVR- or DM-DMPs were significantly related to health care costs. Specifically, an additional year of age increased costs by 1%, the presence of CVD by 43%, a unit increase in the Charlson co-mobidity index by 28%, while an increase of 0.1 in EQ-5D utility decreased health care costs by 7.4%. Moreover, the presence of additional funding in a DMP increased health care costs by 17%, and

patients who were included in CVR- or DM-DMPs had 21% lower costs that patients included in COPD-DMPs. The remaining covariates had no significant association with health care costs.

Model 3, which was the reduced model, fitted the data slightly better than Model 2 because it had a smaller AIC, but it explained two percentage points of variance in costs between DMPs less than Model 2. Another interesting change in this model compared to Model 2 was that additional funding for integrated care became statistically insignificant. However, the p-value of this covariate without using the robust estimator was 0.037, which leaves room for interpreting this covariate as of borderline significance.

Model2-SP shows that employment increased health care costs by 29%, which was as expected because only individuals with a paid job can have productivity loss due to illness absence. Another difference with the results from the health care perspective was that age is no longer significantly associated with health care costs. A similarity between the models from the two different perspectives was that additional funding for integrated care had almost the same effect size and p-value.

5.3.3.2 Results per disease and from a societal perspective.

The results from Model 2-CVR, Model 2-COPD and Model 2-DM are presented in Table 5. The presence of CVD, the Charlson co-morbidity score and the EQ-5D were significantly associated with health care costs in all three models. Additional funding of integrated care was independently associated with costs only in CVR patients. Physical activity had a minor but significant association with costs in COPD. Moreover, employment decreased health care costs by 23% in the COPD sample and was of borderline significance in the DM sample.

5.3.4 Results from descriptive analysis of missings

As Table 6 shows, 17% of the total sample had a missing observation in the physical activity variable and less than 8% in all other covariates included in the analysis. Although the proportion of missing values was relatively low, the cases that had at least one missing observation in one of the covariates included in the analysis (noncomplete cases) seemed to be statistically different from the complete cases. In detail, the noncomplete cases were older, had lower quality of life scores and level of disease management, had less self-efficacy and included more females, more unemployed, more ex-smokers, more people with low educational level, and more singles compared to the complete cases. There were no missing values in costs as we assumed them to be 0 when no health care utilization was reported.

Table 5 Results from the disease specific and from the societal perspective models

	Model 2-CVR (n_p=1006, n_{DMP}=9)		Model 2-COPD (n_p=598, n_{DMP}=4)		Model 2-DM (n_p=302, n_{DMP}=3)	
Fixed effects	exp(β)	p	exp(β)	p	exp(β)	p
Intercept	277.05	<.001	474.23	0.017	120.78	0.099
Physical activity	1.02	0.230	0.98	0.044	1.01	0.815
Employment	0.98	0.839	0.77	0.025	1.26	0.062
Gender	0.95	0.579	1.16	0.163	1.16	0.222
Age	1.01	0.096	1.00	0.633	1.01	0.150
Smoker	1.05	0.713	0.78	0.148	1.07	0.647
ex-Smoker	0.99	0.932	1.05	0.770	1.12	0.344
CVD	1.41	<.001	1.45	0.019	1.47	0.040
Charlson index	1.28	<.001	1.21	<.001	1.28	<.001
Low educational level	1.01	0.936	0.87	0.173	0.95	0.565
Single	0.88	0.196	1.11	0.359	0.99	0.693
EQ-5D	0.14	<.001	0.51	0.009	0.26	<.001
PACIC	0.97	0.518	1.07	0.229	1.11	0.122
Self-efficacy	1.00	0.231	1.00	0.114	1.00	0.442
Additional financing	1.23	0.049	0.63	0.126	-	-
CVR-DM						
Random effects						
σ_e^2 (residual)	1.6246		1.4214		0.7611	
σ_{u0}^2 (intercept)	2.96E-21		4.21E-19		4,1E-19	
ICC	0%		0%		1%	
R_1^2	19%		15%		20%	
R_2^2	76%		65%		83%	
AIC*	3389.67		1939.49		6173.53	

* the AIC is not comparable between these 4 models because the sample population between the models is different

ICC = $\sigma_{u0}^2 / \sigma_{u0}^2 + \sigma_{e0}^2$

Table 6 Descriptive results from the incomplete case analysis

Covariate	n missing	% of N	complete cases mean rank or %	non-complete cases mean rank or %	p value
Physical activity	486	17.0	1179	1217	0.27
PACIC	213	7.5	1337	1286	0.12
Age	176	6.2	1258	1545	0.00
Low educational level	167	5.8	34% low educ. level	55% low educ. level	0.00
Employment	130	4.5	59% unemployed	67% unemployed	0.00
Ex-smoker	91	3.2	51% non-ex-smokers	59% non-ex-smokers	0.00
Smoker	91	3.2	74% non-smoker	72% non-smoker	0.16
EQ-5D	78	2.7	1420	1326	0.03
Gender	76	2.7	44% women	54% women	0.00
Self-efficacy	40	1.4	1437	1352	0.01
Single	24	0.8	74% non-single	64% non-single	0.00

Note: N=2858; Higher mean rank indicates higher scores

5.3.5 Analysis after imputation

Model 2-MI and Model 3-MI were fitted to the imputed dataset (n_p=28547, n_{DMP}=16) as described in section 'Results from the GLMM using complete cases', and the results are presented in Table 7. Age, presence of a CVD, Charlson co-morbidity score, EQ-5D score and being included in a COPD-DMP instead of a CVR- or DM-DMP were significantly and similarly associated to costs as in the complete case analysis. Being an ex-smoker had a positive and almost significant association with costs in Model 2-MI. Additional funding of integrated care was not significantly related to health care costs.

Table 7 Results from the imputed data analysis

	Model 1-MI		Model 2-MI		Model 3-MI	
Fixed effects	exp(β)	p	exp(β)	p	exp(β)	p
Intercept	267.62	<.001	381.35	<.001	363.52	<.001
Physical activity			1.01	0.544		
Employment			0.95	0.442	0.95	0.384
Gender			0.96	0.552	0.94	0.239
Age			1.01	0.015	1.01	0.027
Smoker			1.01	0.908		
ex-Smoker			1.12	0.064		
CVD			1.37	<.001	1.37	<.001
Charlson index			1.30	<.001	1.31	<.001
Low educational level			1.00	0.968		
Single			0.96	0.520		
EQ-5D			0.25	<.001	0.26	<.001
PACIC			1.04	0.216		
Self-efficacy			1.00	0.809	1.00	0.844
Additional financing			1.12	0.201	1.12	0.191
CVR/DM-DMP			0.70	0.001	0.71	0.001
Random effects						
σ^2_ϵ (residual)	1.9818-1.9855		1.6956-1.7058		1.7019-1.7119	
σ^2_{u0} (intercept)	0.1553-0.1554		0.01413-0.015680		0.0207-0.2266	
ICC	7%		1%		1%	
R_1^2			20%		19%	
R_2^2			79-80%		75-77%	
AIC*	9995.37-10118.17		9555.67-9683.95		1939.49	

(n_p=28547, n_{DMP}=16)

5.4 Discussion

In this article, we have developed an appropriate model to incorporate skewed health care cost data in a multi-level analysis by employing a GLMM. Our results indicate that there is large variability in the health care and total costs per patient and the breakdown of these costs into different categories. This variation is present between DMPs but also within one DMP for a particular disease. In bivariate analyses, we found that each covariate was significantly associated with health care costs, except for the patient-perceived level of disease management as measured with the PACIC. The latter is not surprising because patients completed the questionnaire shortly after they had been enrolled into the program.

In the multivariate analysis, we found that being older, the presence of CVD, scoring higher on the Charlson comorbidity index, the presence of additional financing of integrated care and being included in a COPD-DMP instead of a CVR-DMP or DM-DMP was associated with an increase in health care costs, whereas scoring higher on the EQ-5D was associated with a reduction in health care costs. The higher costs in COPD-DMPs can be explained by the severity of the disease itself (reflected in a lower EQ-5D score and lower employment rate) in comparison with CVR and DM instead of differences in the DMPs. These findings were similar for the complete case analyses and the analyses after imputation of missing data, and they confirmed the hypotheses presented in section 'Theoretical model of factors affecting health care costs'. Furthermore, there are no major changes in the effect sizes and significances of these covariates in the reduced model. The covariates employment, gender and self-efficacy contributed to the better fit of the reduced model, although they were not statistically significant in the final reduced model.

Surprisingly, the covariate for age is not significant in any of the disease-specific models, while the same covariate is significant in the overall sample. A possible explanation is that the age of the patients who are enrolled into one of the DMPs for a particular disease is more homogeneous than the age of the total sample. This is confirmed by the smaller coefficients and SEs of age in the three disease samples comparing to the total sample. Another support for this argument is that the statistical power for age is above 97% in all three disease populations, which rejects the possibility of having a nonsignificant age-covariate in the three samples due to small samples.

It is also interesting that physical activity and employment were independently associated with lower health care costs in the COPD sample only. This is probably related to the fact that, in the COPD sample, only 33% of the patients reported to have paid employment, whereas this was 40% and 42% in the diabetes and CVR sample.

The presence of CVD and the Charlson co-morbidity index had a strong positive association with health care costs in each of the disease groups. This was true also in the CVR group, where the Charlson co-morbidity Index was lower because this patient

population contains a considerable proportion of patients at risk of getting a CVD but without diagnosed CVD. This confirms that multi-morbidity is an important health care cost driver [157,170,186].

When the dependent variable is total costs from a societal perspective, employment becomes statistically significant, whereas the independent association between age and costs disappears. This can be explained by the fact that older patients with chronic diseases are less likely to have a paid job [167].

There were few differences in results between the complete case analysis and the analysis after imputation of missing data. Specifically, after multiple imputation, being an ex-smoker had a positive and almost significant association with costs, and additional funding of integrated care was no longer significantly related to health care costs. Although, these are not major differences, MI analysis is more statistically precise than the complete case analysis since the SEs are smaller than in the complete cases. However, imputing observations in a data set derived from a 52% response rate of a survey, as in this case, might be not enough to avoid biases. The relative low response rate is explained by the observational nature of the current study design and is a common phenomenon among cost and evaluation studies of DMPs [152]. Nevertheless, the mean age and percentage females included in each disease sample are similar to the overall Dutch population with the respective diseases.

Considering the predictive power of our models, a 20% value of R_1^2 is in line with the per cent that health care cost models usually predict (i.e. ≤20%) [187]. Perhaps we could have explained more variance in health care costs between patients and DMPs, if clinical data on disease severity would have been included, but these were not available. In addition, we did not expect large differences in estimated mean costs per patient between the DMPs after adjusting for all covariates included in our model. This is confirmed by the R_2^2 of about 85%, which could get even higher if more covariates on organizational-level were included. The 3 months recall period of the patient questionnaire is not considered as a limitation of this study since there is little association between the length of follow-up and the variability in annualized health care costs [187].

The relatively small number of DMPs included in the analysis and the small variability in the type of financing between DMPs are considered as limitations of this study, which may become problematic when sub-group analyses are performed. However, the 16 DMPs are considered to be representative of the DMPs that have been implemented the last 3 years in the Netherlands because (a) they cover all diseases for which DMPs have been implemented, (b) they include DMPs in primary and/or secondary care, (c) they cover a wide variety of different regions and geographic areas with different population density and (d) they vary in disease management interventions and structure of multidisciplinary teams [128]. The study population per disease is also representative of the overall disease population in the Netherlands with respect to age and gender.

Despite these limitations, our study presents reliable results and demonstrates a modelling process of health care costs that can be followed in similar studies in the future. Further research can investigate whether other factors have a relation with health care costs of patients included in DMPs. Such factor could include, on the DMP level, the size of a DMP, the number and type of care providers involved in a DMP and the intensity (i.e. frequency and type of interventions) of DMPs. On patient-level, they may include prior health care utilization, disease perceptions and self-management behaviour. An investigation of cost patterns in patients with and without multi-morbidity focusing on the combination of morbidities could also be an interesting topic for further investigation.

5.5 Conclusions and Policy Implications

There are four main concluding points emerging from this article.

- First, the variability in health care costs of chronic patients within and between DMPs found in this study urges for identifying its causes on patient and organizational-level. This variation means that there is a sub-group of patients with high costs for which the potential cost savings due to DMPs may be higher. The causes of variability should be carefully considered when policies towards integration of chronic care by means of DMPs are being designed.
- Second, care providers and payers (e.g. health insurers) should seriously consider the variability in the target population of a DMP when negotiating about the price of care included in a DMP [47]. For instance, the significantly higher health care costs related to multi-morbidity and/or CVD should be adequately anticipated.
- Third, predictive modelling to identify the 'most suitable' potential enrolees in a DMP can improve the (cost) effectiveness of these programs. For instance, the inclusion of physical activity interventions in COPD-DMPs and the support of COPD patients to get or remain in employment can be potentially linked with reduced health care costs of these patients.
- Last, we found that providing additional funding to support integrated care may increase health care costs at least in the short term, which is in accordance with recent evidence in the Netherlands [93]. This relation should be further investigated and included in the process of redesigning the payment scheme of chronic care.

Appendix 1 Specifying the generalised linear mixed models

In GLMMs the mean of the outcome variable (in our case costs) for the i^{th} patient in the j^{th} DMP is estimated as:

$$E[Y_{ij}|b_i] = \mu_{ij}^{SS}$$

where the expected outcome Y_{ij} is conditional on a latent vector for subject specific deviation from the overall mean of the intercept or slope b_i which equals the expected (subject specific) mean value μ_{ij}^{SS}. The expected mean value is estimated as

$$g(\mu_{ij}^{SS}) = X_{ij}^T\beta + Z_{ij}^T b_i,\ Y_{ij}|b_i \sim F$$

where g is the link function that describes the scale on which covariates in the model are related to the outcome and F is the distribution function for the observed outcome (in this case costs) given b_i. In the fixed part of the GLMM, X_{ij}^T is a covariate vector (including X_0 for the i^{th} patient in the j^{th} DMP) and β is a vector of regression coefficients. The random part, consists of a vector of random effects Z_{ij}^T and the vector for subject-specific deviation b_i and has a normal distribution with zero mean and $q \times q$ covariance matrix G. In other words, the variance of the outcome Y_{ij} is given by

$$COV(Y_{ij}|b_i) = \varphi\, v\, (\mu_{ij}^{SS})$$

where φ is the dispersion (or scale parameter) and v is a known variance function.

Taking these formulas into account, the distribution function, the link function, the covariance matrix, the fixed and random effects should be specified in order to build a GLMM [184,185,188-192]. We followed this process and performed the econometric analysis by using the GLIMMIX procedure in SAS 9.2.

We first specified a saturated model in the fixed part including all available covariates in the analysis and then we specified the random part of the model. Choosing the variance function (v) was the first step in building the model. We chose an unstructured covariance matrix because we specified a random intercepts model (not random slopes), which incorporates only the variance of the intercept [184]. Furthermore, we used marginal likelihood based on Gauss-Hermitte quadrature because the maximum likelihood estimation in the model should express the uncertainty about the b_i, which is unobserved. This maximum likelihood estimation method works well for situations including one or two random effects and two to three clusters [189].

Moreover, the distribution and link function were specified. The literature suggests the gamma, inverse Gaussian, Weibull, lognormal, Poisson and exponential distributions

as possibly suitable distributions for costs, while identity, logarithmic, reciprocal, inverse square, and power are suggested as possible link functions [181,183]. Combinations of these distributions and link functions were made in order to choose the combination that fits the model most adequately to the data. Similar to previous studies [190], we compared the goodness-of-fit of models with the same fixed effects part (i.e. saturated model) and different combinations of distribution and link functions by using the Akaike's information criterion (AIC) and the Bayesian information criterion (BIC).

The results from the combinations of link and distribution functions are presented below. It is clear that the combination of a lognormal distribution and an identity link function had the best fit, because it had the lowest AIC and BIC values. The residual diagnostics (e.g. normal and deviance plots of studentized residuals) were also best for this combination of distribution and link function. Hence, this combination was used in the GLMM analysis.

Goodness-of-Fit tests for combinations of distributions and links

Link	Info criteria	Gaussian	Gamma	Lognormal	Expo
Identity (ID)	AIC	35349.81	-	6181.76	-
	BIC	35364.49	-	6196.44	-
Log	AIC	33798.09	27889.51	6200.43	27901.12
	BIC	33803.77	27904.19	6215.11	27915.03

note: the models with Inverse Gaussian, Poisson, and Weibull distributions did not converge in any instance, thus they are not presented in this table

CHAPTER 6

Identifying and explaining the variability in development and implementation costs of disease management programs in the Netherlands

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Abstract

Background: In the Netherlands, disease management programs (DMPs) are used to treat chronic diseases. Their aim is to improve care and to control the rising expenditures related to chronic diseases. A bundled payment was introduced to facilitate the implementation of DMPs. This payment is an all-inclusive price per patient per year for a pre-specified care package. However, it is unclear to which extent the costs of developing and implementing DMPs are included in this price. Consequently, the organizations providing DMPs bear financial risk because the development and implementation (D&I) costs may be substantial. The aim of this paper is to investigate the variability in and drivers of D&I costs among 22 DMPs and highlight characteristics that impact these.

Methods: The data was analyzed using a mixed methods approach. Descriptive statistical analysis explored the variability in D&I costs as measured by a self-developed costing instrument and investigated the drivers. In addition, qualitative research, including document analysis and interviews, was conducted to explain the possible underlying reasons of cost variability.

Results: The development costs varied from €5,891 to €274,783 and the implementation costs varied from €7,278 to €387,879 across DMPs. Personnel costs were the main component of development. Development costs were strongly correlated with the implementation costs ($\rho = 0.55$), development duration ($\rho = 0.74$), and number of FTEs dedicated DMP development. Organizations with large size and high level of care prior to the implementation of a DMP had relatively low development costs. These findings were in line with the cross-case qualitative comparison where programs with a longer history, more experienced project leadership, previously established ICT systems, and less complex patient populations had lower D&I costs.

Conclusions: There is wide variation in D&I costs of DMPs, which is driven primarily by the duration of the development phase and the staff needed to develop and implement a DMP. These drivers are influenced by the attributes of the DMP, characteristics of the target population, project leadership, and ICT involved. There are indications of economies of scale and economies of scope, which may reduce D&I costs.

6.1 Background

In recent years, the healthcare community has been struggling to identify strategies to better manage the rise in the number of patients with chronic diseases. In the Netherlands, there has been a 17% growth in diagnoses of chronic disease and a 26% growth of patients with multiple chronic disease diagnoses in the past 8 years [193]. One possible method of managing the changes in healthcare that result from the increased diagnosis of chronic diseases is the D&I of disease management programs (DMPs), as has happened in the Netherlands. The Netherlands Organization for Health Research and Development (Nederlandse Organisatie voor Gezondheidsonderzoek en Zorginnovatie (ZonMw)) funded a research project to stimulate the implementation of DMPs and study their impact. Twenty-two healthcare delivery sites were awarded funding to participate in the study by developing and implementing a DMP; the majority of the sites were primary care cooperatives. In exchange for funding and support, each of the sites agreed to participate in research and put a DMP into place. Disease management was defined by the funding organization as:

> a broad programmatic approach to chronic diseases, and a comprehensive care chain of diagnosis, treatment and counseling, as well as prevention, early detection and self-management. The approach is based on multidisciplinary care standards and is organized around the patient and his condition, as much as possible, in conjunction with his surroundings. (Call for proposals, page 7)

In the programs proposed by the care delivery organizations and in the literature about DMPs, patients' participation in the treatment and management of their condition is a key component of DMPs [194], as is the involvement of multiple medical professionals in care planning and delivery [25], and the development and implementation of information systems (most frequently computer-based systems) to support chronic disease treatment and management [195]. The development of the DMPs at the selected sites included interventions altering the existing organization of healthcare delivery (e.g. regular multidisciplinary meetings and regular monitoring of patients) and interventions that were implemented as new processes (e.g. case management, self-management support, ICT). A more detailed description of the interventions is given in elsewhere [128].

In addition to funding for research on DMPs, the Netherlands has implemented a new financing system that impacted care for those with common chronic diseases (an overview of the Dutch healthcare system is provided in Appendix 1). In 2010, a bundled payment scheme was introduced in the Netherlands [47]. Bundled payment is a single payment that covers the multidisciplinary care required by a patient for one particular

chronic disease during a predefined period of time [42]. The aim of this payment scheme was to improve the access, comprehensiveness, continuity, and other aspects of quality of care for chronic patients and to control the increasing expenditures for healthcare for patients with a chronic disease. In the first year of this funding reform, only care for diabetes mellitus (DM), chronic obstructive pulmonary disease (COPD), and cardiovascular risk (CVR) could be contracted in a bundled payment. Under the new payment scheme, chronic care is coordinated by groups of healthcare providers (called 'care groups'). The bundled payment is negotiated between care groups and health insurers and includes 1) the costs of multiple caregivers in primary care (e.g. general practitioners, practice nurses, dieticians, physiotherapists, lifestyle counselor but not medicines, diagnostics and medical devices) as well as 2) the costs of care coordination, 3) information and communication technology (ICT), and 4) professional training and courses for healthcare providers. The latter three groups of costs can be seen as costs for the development and implementation (D&I) of DMPs.

Since DMPs involve a significant reorganization of healthcare delivery, they require substantial development costs (including but not limited to training costs, ICT costs, and costs of redesigning the care delivery process) and implementation costs (such as multidisciplinary team meetings, the costs of coordination between care-givers, the costs of monitoring and feedback). These costs are commonly carried by the organization that implements the program (i.e. care groups). To which extent the D&I costs of DMPs are included in the bundled payment is often unclear. This is despite recommendations to report these costs separately from the healthcare utilization costs and to include them in the price of implementing a DMP [48].

However, some insurers, including the largest one in the Netherlands, are not convinced about the benefits of bundled payment and do not provide this type of funding. Rather, these health insurers provide an add-on payment to cover the D&I costs, whereas the cost of healthcare is funded as before. It is not clear how this add-on payment is defined and to what extent it covers the D&I costs [196]. Considering this uncertainty and taking into account the substantial D&I costs of a DMP, care groups need to be able to correctly anticipate the D&I costs. Failing to do so could be financially disastrous for the providers of DMPs and serves as a disincentive for the implementation of DMPs.

The aim of this paper is to investigate the variability in and drivers of D&I costs among various DMPs and highlight characteristics of the DMPs that may explain the variability in costs during the project period.

6.2 Study setting

The research for this paper was conducted as part of an evaluation of 22 Dutch DMPs spread across different regions of the Netherlands [128]. The DMPs were categorized in CVR (n=9), COPD (n=4), DM (n=3), mental diseases (n=3), and other (n=3). The 'other' disease category includes DMPs for stroke, heart failure, and mix of CVR, COPD and DM. The Ethics Board of Erasmus University approved the data collection. All content has been anonymized.

6.3 Methods

We used a mixed-methods approach to analyze data on D&I costs. To this end, we used descriptive statistical analysis to explore the variability in D&I costs, as well document analysis and interviews with project leaders, managers, and professional care givers.

6.3.1 Quantitative methods

All development and implementation costs associated with the 22 DMPs were systematically collected. We developed a template that was based on the CostIt instrument of the World Health Organization (WHO) [197]. This template was completed during face-to-face interviews with DMP managers. During these interviews we also asked managers whether they had additional financing to cover the specific elements of disease management. The development costs included all costs made during the preparation phase of DMPs, e.g. labor costs for brainstorming sessions, training costs, and ICT support costs. The implementation costs included costs of multidisciplinary team meetings, coordination between care-givers, monitoring and feedback that occurred the year after the DMP implementation. We collected the development and implementation costs regardless of the budget holder for their financing; the budget holders could include care groups, health insurers, and/or government. The labor costs were calculated using the full-time equivalents (FTEs), duration of involvement in the project and the gross salary of medical, administrative, ICT, management and other personnel. Operating costs (including costs of professional courses, information/ communication, licenses, and materials) were calculated based on volumes and unit prices as stated in the template. Capital costs (such as building and purchase of ICT) were calculated based on their volume (for buildings that was square meters) and unit prices (for buildings that was Euro per square meter) and they were amortized over their lifespan as suggested by the WHO [197]. In the analysis, we included the development costs during the development phase, the annualized development costs, and the implementation costs in the year after implementation.

In addition to D&I costs, we also collected data about the duration of the development phase (in months), the number of patients participating in a DMP, the total FTEs available to the organization providing a DMP, and the FTEs dedicated to developing and implementing the DMP. The level of chronic care integration was also measured at the start of providing a DMP and a year later by using the Dutch translation of the Patient Assessment of Chronic Illness Care (PACIC) [168]. This questionnaire was distributed to participants of 19 DMPs (no data for the 3 mental disease DMPs was available). The mean PACIC value of the participants in each DMP was used in the analysis.

Descriptive statistics were used to investigate the variability in D&I costs among 22 DMPs. Pearson correlation coefficients and Spearman correlation coefficients were calculated for normally distributed and non-normally distributed variables, respectively. The normality was tested based on the Kolmogorov-Smirnoff test. We also performed an analysis of variance based on ANOVA and Kruskal-Wallis estimates to explore differences in the development and implementation costs among disease categories. We also performed an analysis of variance to investigate differences in D&I costs among different payment methods during the development and implementation phases. The payments were categorized in normal (e.g. for GPs this is a mixture of fee-for-service and capitation payment), normal plus add-on payment for D&I costs, and bundled payment. Considering the small number of observations (n=22) we also looked into various associations using scatter plots and graphs.

6.3.2 Qualitative methods

In order to understand how various characteristics may influence the costs associated with the D&I of DMPs, we examined how program plans 'travel' from the grant proposal to the D&I of the DMPs [9], as well as what actually happened during the D&I phases of the program by exploring and analyzing the multiplicity of D&I in practice [198]. This approach enabled us to gain a deeper knowledge of the activities implicit in DMPs, including activities that influence how programs develop, how programs use the provided finances, and how project teams overcame (or not) difficulties in the early stages of programs.

Document analysis was the first step of the qualitative data collection. The documents analyzed included the grant applications and project plans submitted by project leaders, the call for proposals (Diseasemanagement chronische ziekten), and care organization websites. The documents were analyzed inductively to gain a better understanding of the DMPs, project leaders, and care providers. The content of the documents informed the development of the interview guide, which focused on D&I of DMPs in practice.

Two in-depth case studies were selected for this paper, which highlight the different D&I costs in the DMPs; 15 interviews with project leaders and clinicians were conducted and used in the case studies presented in this manuscript. Questions about the history

and contexts of the DMPs were asked. Interviews were digitally recorded and detailed notes and observations were also made during the interviews. Interviews were conducted in Dutch or English and ranged from 30 minutes to 90 minutes. Interviews were transcribed and coded into themes. Quotes were translated by a native English speaker; the translations were later confirmed by a native Dutch speaker.

To better understand the variability in costs from a mixed methods perspective, the primary economic researcher (AT) and the primary qualitative researcher (BHW) met regularly and jointly reviewed the data. The economic and qualitative data have been integrated iteratively, after consensus by all authors. This was done through frequent meetings between the first two authors and the rest of the authorship team.

6.3.3 Ethics statement

The study protocol was approved by the ethics committee of the Erasmus University Medical Centre of Rotterdam (September 2009). For more details see Lemmens et al., 2011 [128].

6.4 Results

Adequate understanding of complex policy structures and the impact of their change requires multiple types of information. Mixed-methods research facilitates this by combining qualitative and quantitative research methods in order to identify, decompose, analyze, and understand complexities in healthcare [199]. Our research found a large variability in D&I costs between the researched DMPs. We uncovered three common characteristics of the studied DMPs that may explain the variability in costs between programs. These characteristics include attributes of the interventions, ICT systems, and the experience of the project leaders. The history of the programs, including personnel time invested and ICT systems already in place, may also play an important role in the variability in costs and is an underlying characteristic.

6.4.1 Variability in D&I costs and cost drivers

As Figure 1 shows, the development costs varied from €5,891 to €274,783 across DMPs and the implementation costs varied from €7,278 to €387,879 across DMPs. There was also large variation in D&I costs across DMPs in the same disease category. In some cases the development costs were higher than the implementation costs and in some other cases not. In addition, the four DMPs with the highest development costs among all DMPs also had the highest implementation costs. When annualized, the development costs varied also largely between and within disease categories and were also positively associated with the implementation costs.

Figure 1 Development and implementation costs per DMP

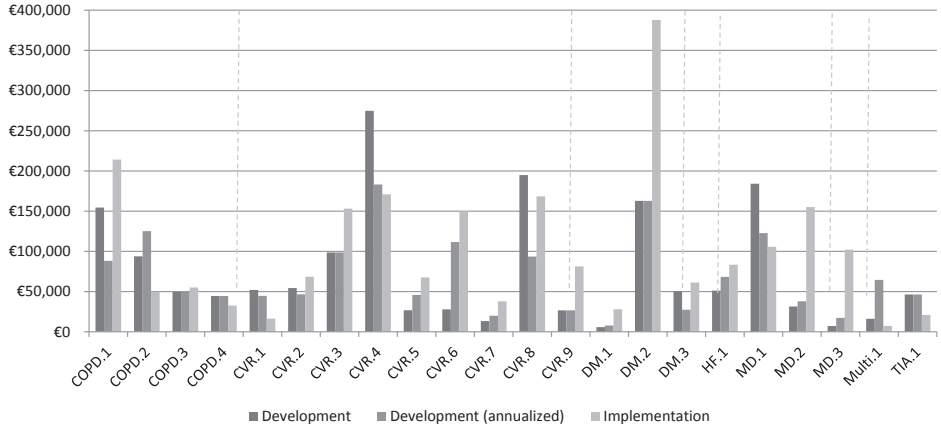
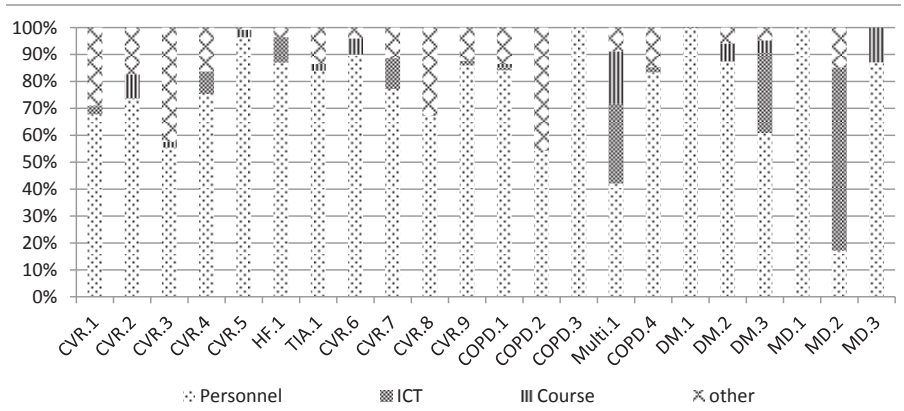


Figure 2 Share of total development costs per cost component



Personnel costs were the primary component of development costs across 18 of 22 DMPs, accounting for more than 60% of total development costs (Figure 2). They were followed by ICT costs (maintenance and licensing) and the costs of professional courses as the main cost components of development costs.

The results from the descriptive statistical analysis are presented in Table 1. This table shows that our sample consisted of DMPs varying in the duration of the development phase (range: 3; 25 months), number of patients participating in the DMP (range 75; 3,400), total number of FTEs in an organization (range 1; 2,850), and number of FTEs involved in developing a DMP (range: 0.1; 2.5). The mean development costs were €75,832, the mean annualized development costs were €69,749 and the mean implementation costs were €100,827 across all 22 DMPs. The mean PACIC at implementation was 2.88 and a year later was 2.95. The Kolmogorov-Smirnoff test showed that the variables 'number

Table 1 Descriptive statistics

	Mean	SD	Median	Min	Max	IQR
Development duration (months)	12	6	12	3	25	6
Patients participating in DMP[#]	801	986	300	75	3,400	957
Organization FTEs[#]	433	841	33	1	2,850	256
DMP FTEs	0.76	0.58	0.60	0.10	2.50	0.63
Development costs[#]	75,832	72,727	49,972	5,891	274,783	85,917
Annualized development costs	69,749	47,807	48,141	7,855	198,188	66,704
Implementation costs	100,827	86,776	74,836	7,278	387,879	117,079
PACIC at baseline[#] (1-5 best)	2.88	0.29	2.81	2.25	3.60	0.35
PACIC at year 1[#] (1-5 best)	2.95	0.28	2.99	2.44	3.62	0.40

[#] The Kolmogorov-Smirnoff test rejected the assumption of normally distributed data; SD: standard deviation; min: minimum; max: maximum; IQR: interquartile range (Quartile 3-Quartile 1); FTE: full-time equivalent; implementation costs accrued within the first calendar year of DMP implementation

of DMP participants, 'organization FTEs', 'development costs' and 'PACIC at implementation' and 'PACIC a year later' were not normally distributed.

The correlations between the D&I costs and potential cost drivers are presented in Table 2. Total development costs were strongly correlated with implementation costs (ρ=0.55), development duration (ρ=0.74), and number of FTEs dedicated to the development of a DMP (ρ=0.54). The latter was found to be correlated also with the development duration (ρ=0.49). The annualized development costs were correlated with the implementation costs (ρ=0.65) and number of FTEs for the development of a DMP (ρ=0.52). The results also showed a negative correlation between PACIC a year after implementation and development (ρ=-0.27) and implementation (ρ=-0.24) costs.

The relation between development costs and the total number of FTEs in the organization that provides a DMP is illustrated in Figure 3. This figure shows that large orga-

Figure 3 Association between development costs and total number of FTEs in the organization

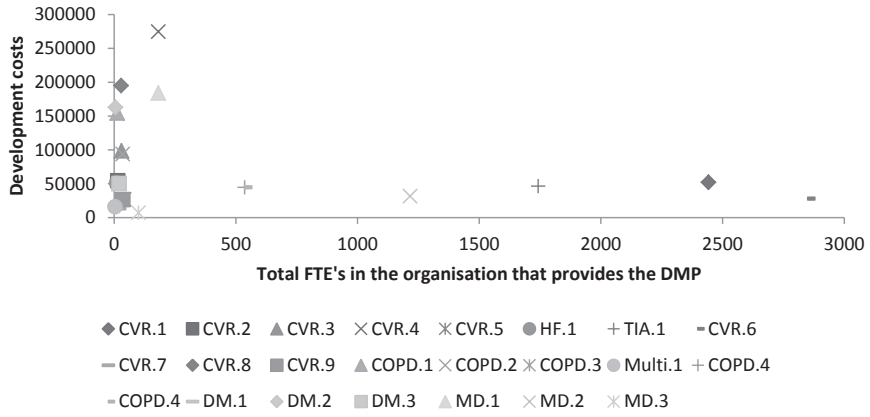
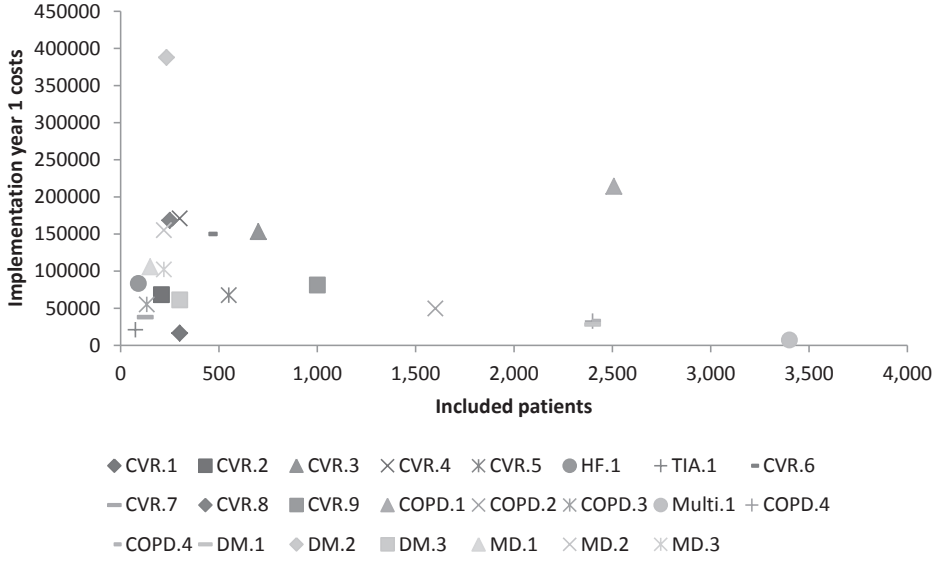


Table 2 Correlation coefficients

	Development costs	Annualized development costs	Implementation costs	Development duration	DMP participants	Organization FTE's	DMP FTE's	PACIC baseline	PACIC year 1
Development costs	1								
Annualized development costs	0.79 (0.000)	1							
Implementation costs	0.55 (0.008)	0.65 (0.001) [#]	1						
Development duration	0.74 (0.000)	0.24 (0.284) [#]	0.27 (0.228)[#]	1					
DMP participants	-0.12 (0.600)	0.02 (0.922)	-0.08 (0.707)	-0.09 (0.688)	1				
Organization FTE's	-0.03 (0.887)	0.02 (0.940)	-0.00 (0.988)	-0.03 (0.880)	-0.14 (0.549)	1			
DMP FTE's	0.54 (0.010)	0.52 (0.013) [#]	0.16 (0.482)[#]	0.49 (0.022)[#]	-0.04 (0.869)	-0.35 (0.110)	1		
PACIC baseline	-0.22 (0.366)	-0.02 (0.937)	-0.21 (0.388)	-0.24 (0.323)	0.21 (0.380)	-0.40 (0.095)	-0.29 (0.232)	1	
PACIC year 1	-0.27 (0.051)	-0.08 (0.049)	-0.24 (0.044)	-0.21 (0.396)	-0.23 (0.350)	-0.02 (0.932)	-0.28 (0.909)	0.64 (0.003)	1

[#] Based on Pearson correlations; FTE: full-time equivalent; in brackets are the p values of the correlation

Figure 4 Association between implementation costs and number of patients participating in a DMP



nizations had relatively low development costs compared to small organizations. This relation remained between the annualized development costs and the total number of FTEs in the organization that provides a DMP (see Figure 4 in Appendix 2).

Figure 4 illustrates the relation between implementation costs and the number of patients participating in a DMP. We see that there might be a small negative relation between the two variables.

The results of the analysis of variance of D&I costs comparing different payment methods as well as different disease categories were not statistically significant. Figures illustrating these variances are presented in Appendix 2.

6.4.2 Characteristics of the disease management programs

While each of the practice sites had a different method of addressing chronic disease through the funded DMP, the qualitative research uncovered characteristics that help to understand the differences in D&I costs. These characteristics include attributes of the DMP, ICT systems, and the experience of the project leadership teams. Qualitative data from two case studies will illustrate these characteristics. The diabetes case (DMP number 17) had relatively low D&I costs, while the CVRM case (DMP number 10) had high D&I costs.

6.4.2.1 Case study: Diabetes (DMP number 17)

One of the project sites with a focus on adult-onset diabetes developed a DMP for clinicians and patients. The clinicians and project leaders at the site have invested time and

effort in improving diabetes care for patients since 1999 by working in cooperation with the hospital and specialists, developing care protocols, and contracting with providers. Around 2006, the changes in diabetes care were formalised into an early form of disease management. The studied program was a continuation of a previously funded program. This site had two project leaders, the first of which was hired for the previously funded program to assist with the development of an electronic medical record and stayed on through most of the funded DMP. The first project leader was a professional healthcare consultant with a background in economics. The second (and current) project leader was the assistant of the first and took over the project leadership when the first project leader left, after a period of mentored transition.

Program attributes

For the disease management study, the project leaders worked with healthcare professionals to change how they thought about care and creating multidisciplinary care teams, implemented an updated networked electronic medical record system with a patient portal, and oversaw self-management education for patients.

The multidisciplinary care teams included nurse specialists, dieticians, general practitioners, nurses, chronic disease specialist assistants, ophthalmologists, podiatrists, and/or internists; members of the care team were located in multiple GP and specialist offices throughout the region. The care team worked together, communicating with each other frequently.

> *I think that working closely with the dietitian especially... We sit together, we discuss a lot, and we can call the GP or practice nurse, and yes, several people are looking [at the case].* (Interview with diabetes specialist nurse)

Communication was seen as key to coordinating care. It was common for the staff, such as doctors, nurses, dieticians, and other clinical professionals, of disease management teams to sit together in a shared office and communicate about patients and care; however, this was not the only way that communication happened. The diabetes program had regular meetings with clinical professionals from multiples GP offices with the project leader overseeing the meetings. The project leader also sent out emails, reports, and posted information about the DMP online. Clinicians also communicated with one another through the networked electronic medical record. This system of coordinated care has been developed by investing in time for the project leader and clinician stakeholders over the years of the program.

One of the efforts of this disease management project focused on self-management through educating patients about diabetes. There were a variety of formal and informal educational opportunities for patients: group classes, clinical visits, and online. Patients could participate in voluntary classes, which were led by a nurse, doctor, and/or non-physician chronic disease specialist assistant. Clinicians were trained to lead by experts

in diabetes education, an additional D&I cost-item which was hoped to be recouped in a reduced need for individual education sessions with patients. However, the classes were no longer offered as a result of lack of patient interest and attendance.

Internet-based communication systems

For the diabetes project, as is the case for many medical practices, the full development and use of an electronic medical record (EMR) was a complicated process. The GP offices involved in the diabetes project began working with software developers in 2006 to develop a more-limited version of an EMR. The project leaders, especially the first project leader, worked with clinicians and software developers to enhance the record. The newly enhanced record allowed for viewing the record by multiple clinicians (and the project leaders) at different physical locations, electronic referrals, and messaging between clinicians and between clinicians and patients.

Internet-based communication (ICT) systems, such as the EMR, have been seen as a remedy for the many predicaments in healthcare delivery and quality, one that promises more than is realized in most cases [200]. The implementation of ICT systems in healthcare delivery has been a lengthy process for project leaders and clinicians involved in the DMP, requiring planning, developing, implementing, and tailoring the system before the system can begin to meet the needs of clinicians and patients. At this site, the development of the ICT system began well before the program was funded as part of the study. By working with a previously developed system, this project leader and leadership team had the opportunity to gain the needed support from stakeholders, as well as work through the inevitable bugs and challenges in the system before the funded DMP officially began. These challenges included:

> *The fact that the software builder couldn't deliver what they said they would deliver. And still now we do not really have the perfect system and the perfect system does not exist, I know. But there are too many things that we want. But, there is no other software builder at this moment that DOES have it. The software builder itself has been in bankruptcy 2 times, once in 2007 and 2 months ago for the second time. Fortunately they worked together with another department who had been able to go on with the system, so that we were not cut off.* (Interview 1 with project leader 1)

As noted in the quote above, the investment of staff time and the hurdles that frequently occur in the early development phases of a networked electronic medical record occurred, in part, before the study and in the early phases of the study. Since the computer program had already been chosen, the time and effort needed to select a computer program and coordinate the program with existing record systems were not part of the study. As a result, much of the development work and growing pains of implementing an ICT system were not seen in the D&I costs for this aspect of the DMP.

One of the goals of the funded portion of the DMP was the D&I of a patient portal for the networked electronic medical record. While the project team was able to develop a patient portal in a timely manner, the development of the portal does not guarantee usage.

> *Yes, we do have that but no-one uses it. We have talked and talked to get people to look, [telling patients] you have your own care dossier, your own plan, we can agree on goals there, you can report on how it is going, you can also tell us what does not go well, or if you have questions. Really easy, you can do that from your chair at home, you don't have to come here if you don't want to. But people don't want that. It has cost money, because in order to offer the portal we had to expand our software package. Of the current 2700 people with diabetes I believe 15 now have a care plan.* (Interview 1 with second project leader, diabetes project)

While rates of patient participation in the portal were low, the numbers were expected to increase in the future as more internet-friendly patients are diagnosed. Though the usage remains low, this D&I cost is expected to have lasting impact well after the project period.

The networked electronic medical record has travelled from plan to action successfully in principle, in that the portal was developed in a timely and cost-effective manner, but not in the current day-to-day reality of the program, as very few patients used the portal at the time of the interview. This investment in future patients via a patient portal can be seen as a D&I cost. While the patient portal was successfully developed from a technical point of view, the implementation in practice will still require much effort on the part of the clinician, the project leader, and the expected future patients, who too must learn how to use the new system and whose time is frequently overlooked in accountings of D&I costs.

Project leadership

The cooperation hired an experienced project leader to oversee the diabetes DMP. Much like the groundwork done with the ICT system, the hiring of this first project leader occurred before the funding of the study. The project leader saw her role as giving support, both material and strategic:

> *And I think that a unique thing is that what I do is I'm able to give support on the strategic thinking… where do you want to go to, what are the goals, what's your mission, what's the gain of it. And the second is how can we achieve that. So it's not a consultant with only the advice but also what does it take to get there.* (Interview 1 with project leader 1)

The first and second project leaders led meetings, created reports from data extracted from the networked electronic medical record, and coordinated the efforts of the study team, such as sending out surveys. When the project leader resigned, she passed the role on to her assistant.

Effective leadership is crucial for bringing projects to life. In fact, in their seminal article on DMPs, Wagner et al. (2001) point out that making the change needed for DMPs is "difficult, if not impossible, without strong leadership" [25]. Leadership support was (and, in general, is) needed for multiple aspects of the DMP: for the successful implementation of a health ICT system [201], to guide the vision of the improvements in chronic care treatment and management [35,194], and to facilitate change in the healthcare delivery [202]. Yet project leadership skills and efficiency grow over time, as the project leader gains the trust of the clinicians, as the project leader is better able to understand the needs of the clinicians and patients, and as the project leader and the clinicians are able to adapt to one another.

6.4.2.2 Case study: Risk of cardiovascular disease (DMP number 10)

A DMP conducted in two GP offices focused on improving care for those with an elevated risk of cardiovascular disease. The project team consisted of two GP-researchers (one of which served as a part-time project leader) and a nurse manager, who did the day-to-management of the program and study. As written in the grant proposal, the key elements of the program were*:*

a) a patient choice program to promote a commitment to the formulated treatment goals
b) a focus on reaching people with a low socio-economic status (SES)
c) the use of a web-based patient record (Grant proposal)

This DMP was a newly formed project, developed by the project leader who had recently completed a Masters in Healthcare Management from a nearby university.

Program attributes

The disease management project focused on providing coordinated care with multiple clinicians to a challenging population: patients with an elevated cardiovascular risk and a lower SES.

> *We have many patients, about 20% of the patients in the GPs practices are known to the GP as having one form of elevated cardiovascular risk. That's a very big number of patients. Of those patients, about 8 or 9% are under regular control of the GP. And from those, a small part has a low SES. Especially patients at low SES do not follow our advice; you can see that as you look at the numbers. Most people, more people at low SES, dying of cardiovascular diseases, more people smoking… That's the most important start of our project. And we don't reach people with low SES, so we are looking at new methods of treatment of people with low SES.* (Interview 1 with project leader)

The patients with a lower SES were, and commonly are in healthcare, seen as a tricky population with multiple problems, less access to resources, and lower rates of literacy. Providing care and self-management education to this population was expected to be (and was) challenging for the project leadership team and clinicians, requiring a signifi-

cant time investment. To be overcome, these challenges required effort on the part of the clinicians, patients, and project leaders.

Much of the investment of time for project leaders, in general, comes in the early stages of the DMPs. As time goes on in the course of programs, project leaders develop a better sense of the population and are better able to tailor attributes of the DMP for the needs of specific populations. This was the case in the CVRM program, as the project leader noted below:

> *Then we ask the patient, do you want to look at your own patient file on the internet? And when he says yes, he can open his own file and see his own cardiovascular risk profile on the internet. Because we suppose that not every patient with low SES has a computer at home or can look at his file on the internet. So that's why we ask this to every patient.* (Interview 1 with project leader)

This patient population required (more) time and effort from project leaders, clinicians, and medical office staff, as the patient population may not have had internet access, may have spoken limited Dutch, and may have had fewer economic and social resources for support. Accommodating this population to ensure good care required time during the clinical visit and, for the project leaders, time during the D&I of the DMP.

However, the challenges with the patient population were not the only challenges that the project leadership faced.

> *Well, you've got to separate the problems: content level and organizational level. Content, I think, it actually runs smoothly. We must, of course, continue to develop, but that is going the way we want. Organizationally we have some problems. [Primary care] practices are (…) very large organizations now. So before we begin, we have to convince everyone of the importance of the research. That takes a lot of effort. Plus the implementation of such a project, in practice is not simple because practices are large organizations where 30 people work. Plus there are other members of the care group that need to be involved: the physiotherapist, the dietician.* (Interview with the project leadership team)

Effort and accommodation, in the form of meetings, telephone support, and emails, was needed to assist the clinicians in implementing the changes needed for implementing a DMP and for conducting research on the program.

Internet-based communication systems

As was seen in many other projects, the D&I of the networked electronic medical record required much time and effort over the course of years; this effort included working with outside vendors, outside educators, and outside funding agencies, as well as working with clinicians and GP office staff. The effort did not stop; changes and further tailoring continued after the record was in place. The project leaders and manager were key in these activities.

> *And for the development of cardiovascular risk management, this is how far we are now: we have funding. We are now working with contract negotiations. And then we can start developing and the ICT supplier, if they are fast, can get us a beta version in three months' time. We hope that we can really start with ICT in March, February… well, of course it is a problem to get financing. A negotiation problem. Yes, but we are happy that we have had luck.* (Interview with the project leadership team)

Because health insurers provided some of the financing needed for the D&I of the networked electronic medical record, there was much coordination work needed. Health insurers required extensive plans, budgets and presentations before financing was awarded. This was in addition to the work needed to develop the record, such as working with the developers and clinicians. Patient portals were included in the development of the networked electronic medical record, allowing patients to go online and access their record.

> *You can see here, patients with active risk. This is what patients can see at home. The treatment goal of this patient was weight reduction of 6 kg in 3 months. And you can see at this point, he has reached a risk reduction of 80% of his goal. … Here is the plan and what he or she still has to do is treating hypertension and becoming more adherent for medication. But this patient has chosen for weight reduction in a first step for cardiovascular risk treatment.* (Interview 1 with project leader)

While project leaders hoped that the implementation of clinical information systems would improve care for those with a chronic illness, the D&I of the system to this point has come at a significant time-cost in both sites. As Wears and Berg noted, while electronic medical records are often thought of as a panacea for the ills of medical documentation, this is often more dream than reality [200]. To meet the goal of including patients into the patient portal of the new record system, additional time on the part of the clinician and of the project leader was needed, time to tinker with the system, to tailor the system to the needs of the clinicians and of the individual patient, who may or may not have had computer access.

Project leadership

For this two-practice project, project leadership took the form of a team of two GPs and a nurse manager. The nurse's duties included interacting directly with staff at the practices, coordinating the research efforts, and aiding the practice staff as they adopted disease management principles. The work of the project leadership team started:

> *by organizing meetings. That's why we start with 4 meetings and why we start at practice level. Speak with the GPs and the nurses. And we have learned to start slowly, go slowly. I will not tell my GPs to start with 100 patients but will tell my GPs we will start very slowly. … But if it works, we have to change the practice. I hope it works. But we have to wait for it still.* (Interview 1 with project leader)

Changing the practice was seen to need to begin slowly in order to gain support from clinicians and staff at the two GP offices. The project leader saw this coordination and background work as necessary before large-scale changes in patient care were implemented. While this work can be seen as an investment, it was likely a notable source of D&I costs.

Another significant challenge for the project leadership team was to work to procure additional funding for the DMP.

> *Step by step, we write down [the plan] for the insurance company. Because they will first look at the plan. After they will decide whether to give us some money. And when you are going to visit your GP, the GP will receive 9 Euros. But when you do the visit according to our rules of cardiovascular risk management, we think that the consult will take half an hour so we have asked to the insurance company not 9 Euros but 25 Euros.* (Interview 1 with project leader)

The complete implementation of the program goals cost more time at the patient care level as well as the time and effort invested at the project leader level to procure more funding.

6.5 Discussion

The findings of this study show that large variation exists in the D&I costs of DMPs implemented in the Netherlands. This variation can be explained by the large variability in DMP development duration, size of DMP providing organization, and the level of care in the providing organization prior to the implementation of a DMP. The qualitative analysis showed that these characteristics were associated with the attributes of the interventions, project leadership, and the history of the ICT systems used in a DMP.

The DMP development duration is positively related to the labor intensiveness during the development phase and development costs. Considering that the development costs are highly positively correlated to the implementation costs, the length of the development phase is an important cost driver of D&I costs.

The research on the case studies and other qualitative research conducted in the remaining 19 sites highlighted that the D&I of an ICT system was an involved process. While previous literature shows that a well-developed ICT system is one of the main preconditions of successful implementation of bundled payments and DMPs in the Netherlands [47], the work required to develop and implement ICT systems was, at the sites, time-consuming and costly. In the diabetes case study, the majority of the development work involved in implementing the ICT system occurred before the study period, but nonetheless the work did happen; however, the cost for this work is not included in the financial data in the diabetes project. As the ICT work (and the costs associated) was included in the D&I period of the CVRM project, this may be an explanation for variation

in costs. The D&I of adequate ICT is important for all stakeholders in chronic care. It is important for care groups with the aim of achieving lower D&I costs, for health insurers in order to contract chronic care at lower cost, and for public authorities with the purpose of controlling healthcare expenditure by supporting managed care for chronic diseases. As our data shows, this D&I requires time, financial support, and a flexibility of goals, targets, and timelines, no matter when it occurs or how the D&I is funded.

The qualitative research conducted at the sites revealed that the role of the project leader was an important one, with more established projects with experienced project leaders and managers spending less time on the early development of the programs. Project leaders were responsible for guiding the programs, working with clinicians, delegating responsibilities, and developing contacts with outside funders and vendors. In the studied sites, we saw that in projects with a longer history (and with a project leader with more experience in leading healthcare projects and in the DMP project in specific), the relationships needed for smooth, efficient project management were likely developed in the early years of the programs and the costs for these efforts have not been included in the D&I costs (as was seen in diabetes project). In the CVRM project, these relationships were in the process of being developed during the study in an incremental manner through meetings, developing project plans, and the slow introduction of changes. Project leadership, in general, was especially relevant in that organizational and management failure threaten the successful implementation of disease managed care facilitated by bundled payment in the Netherlands [47].

Project leaders had a fluid role and flexibility within the project, as meeting project goals often requires adaptation. Whether by offering new tools online or printing for patients who have limited computer access at home, this constant adaptation by project leaders and clinicians can be seen as "persistent tinkering in a world full of complex ambivalence and shifting tensions" [203]. Through tinkering, project leaders worked to meet the changing needs of patients, of the healthcare system, and of themselves. Yet tinkering was a slow and often invisible process, as was much of the work of project leaders when tailoring interventions, applying for funding, or working with researchers. This tinkering was constant during the study and programs, but as our data shows, appeared to be more prevalent in the D&I stages as the project leaders are working with new vendors, systems, and care plans. This prevalence of tinkering in the early stages of a DMP may have resulted in higher D&I costs.

The specific DMP populations, too, may have had a significant impact on the D&I of the DMPs. The CVRM DMP was working with patients with low SES, many of whom were reported to be complex patients with limited access to resources. Accommodating the needs of this population may have required more tinkering, more effort from project leaders, and more time from clinicians. These characteristics could have played a role in the higher D&I costs for this site.

Our findings also suggest that large organizations providing DMPs are more likely to have lower D&I costs than smaller organizations. This indicates the existence of economies of scope where large organizations may have already established ICT systems, managerial knowledge, and available capital in other care (e.g. public health and prevention) and disease areas that can be also used in the development of disease specific DMPs. This is supported also by the negative relation between the existing level of disease managed care (as measured by the PACIC) in the first phase of implementing a DMP with D&I costs. This unveils existing synergies between projects within organizations. The economies of scope may appeal financially attractive to DMP providers since they might increase the profit margin of providing DMPs for different diseases. The provision of DMPs that could address different disease areas and multi-morbidity could also tackle the criticism of the current DMPs that they are narrowly focused on a single disease while chronic patients need broader care because they often have one or more other diseases (55% of the patients in our sample have more than one chronic disease). Such a development could also tackle the hesitations of health insurers in contracting DMPs.

Moreover, the minor, though negative, relation between the number of DMP participants and the implementation costs, as illustrated in Figure 4, may indicate the presence of economies of scale. The more patients included in a DMP, the lower the marginal costs of implementation. This can be attributed to fixed costs that are divided by more DMP participants. Capital and operating costs, which are included in the implementation costs, are known cost components subject to economies of scale. This financial advantage of large organizations may attract health insurers to purchase DMP from them hoping for a lower bundled payment per DMP participant. However, as in all industries, the number of participants that lowers the marginal costs of DMP implementation should be investigated because further inclusion can lead to higher costs.

Furthermore, we found no evidence of relation between D&I costs and DMP payment method. Similar to case two, many DMPs reported challenges to get additional financing for the provision of a DMP. However, this did not lead in all cases to higher D&I costs. A previous study found a positive relation between additional funding for disease managed care and healthcare utilization costs [204]. Therefore, care groups should be careful in setting the prices of DMPs when negotiating a bundled payment because that price should cover not only the costs of healthcare for the particular disease but also the D&I costs.

There was also no relation found between D&I costs and type of disease addressed by a DMP. That suggests that none of the diseases studied here can be characterized as "cherries" or "lemons" in the chronic care market with respect to D&I costs. This fact may enable the broadening of the scope of diseases that a DMP addresses by making every disease equally financial attractive to care groups.

The 22 DMPs are considered to be representative of the DMPs that have been implemented the last 3 years in the Netherlands because (a) they cover all diseases for which DMPs have been implemented, (b) they include DMPs in primary and/or secondary care (the most common settings for DMPs), (c) they cover a wide variety of diverse regions and geographic areas with different population density and (d) they differ in the attributes of the DMPs put into place and in the structure of multidisciplinary teams [128]. The study population per disease is also representative of the overall disease population in the Netherlands with respect to age and gender.

The findings of this study are relevant to primary care practices in the Netherlands as well as to health policy makers and primary care practices in other European countries that have implemented or are planning to implement DMPs to achieve integration of chronic care. The programs in this study represent a diversity of chronic diseases that can be addressed by DMPs, ranging from common chronic diseases such as diabetes, CVRM, and COPD, to less frequently addressed chronic diseases such as depression, eating disorders, and mental illnesses. The programs, while diverse, had features in common with other DMPs outside of the Netherlands: addressing the issues of chronic illness through coordinated care, through the use of ICT systems, and through the promotion and implementation of self-management education. This research also provides unique insights into the role of project leaders and of the impact of the history of the programs on D&I costs. Policy makers, DMP designers, and primary care practices in the Netherlands and in Europe can explore the possibilities to contain D&I costs at a minimum level by enhancing leadership and ICT in DMPs as well as exploit existing economies of scope and economies of scale in the provision of DMPs.

6.6 Conclusions

The conclusions of this paper can be summarized into:

- There is wide variation in D&I costs of DMPs, which is driven primarily by the duration of the development phase and the labor intensiveness needed to develop and implement a DMP.
- The level of disease managed care in an organization prior to the provision of a DMP is negatively associated with the D&I costs of this DMP.
- Assisting care groups to develop adequate ICT systems for disease managed care is a win-win situation for all stakeholders.
- It is crucial to define the right mix of DMP interventions and target population and to incorporate these mixes in the planning and budgeting of the DMP development phase.

- There are indications of existence of economies of scale and economies of scope, which may reduce D&I costs. Care groups and health insurers should explore the potentials in exploiting them in a mutually benefiting manner.
- The work done before the sites are awarded study funding, especially in relation to ICT systems, saves time and money during the program and study.
- The experience of project leaders may play a fundamental role in the development and early intervention efforts of the DMP.
- Programs with a longer history, more experienced project leadership, previously established ICT systems, and less complex patient populations had lower D&I costs

Note

Some of the results presented in this paper have been previously published in:

Cramm, J.M., Tsiachristas, A., Hipple-Walters, B., Adams, S., Bal, R., Huijsman, R ., Rutten-Van Mölken, M.P.M.H., Nieboer, A.P. (2013). The management of cardiovascular disease in the Netherlands: analysis of different programmes. International Journal for Integrated Care. 13:e028. Impact Factor of 1.75/SE2.

Cramm, J.M., Tsiachristas, A., Adams, S.A., Hipple-Walters, B.H., Bal, R., Huijsman, R., Rutten-Van Mölken, M.P.M.H., Nieboer, A.P. (2014). Evaluating Disease Management Programmes in the Netherlands. Sociaal-Medische Wetenschappen (SMW).

Cramm, J.M., Tsiachristas, A., Hipple-Walters, B., Adams, S., Bal, R., Huijsman, R., Rutten-Van Mölken, M.P.M.H., Nieboer, A.P. (2011). Evaluation cardiovascular disease management programmes: preliminary results. Den Haag, NL: ZonMW.

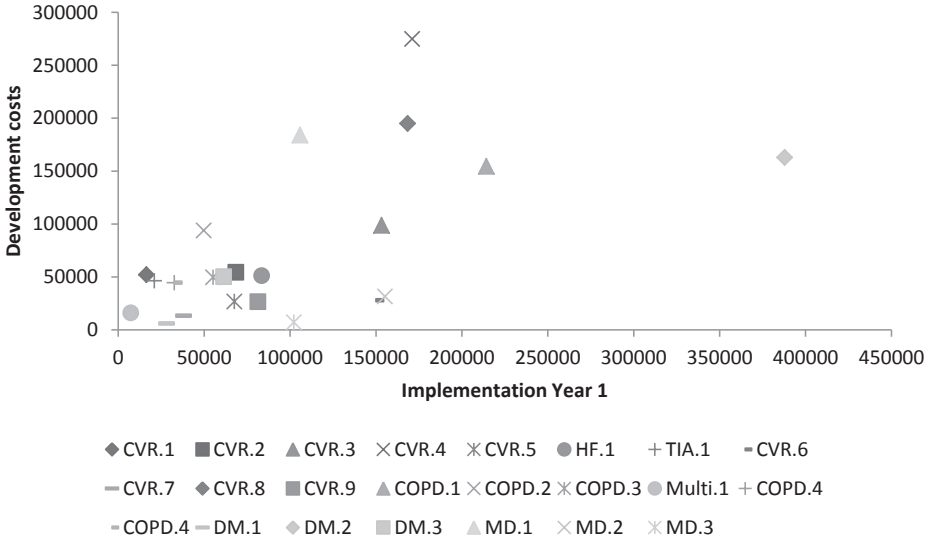
Appendix 1 Background information about the Dutch health care system

Undoubtedly the dominant issue in the Dutch health care system at present is the fundamental reform that came into effect in 2006. With the introduction of a single compulsory health insurance scheme, the dual system of public and private insurance for curative care became history. Managed competition for providers and insurers became a major driver in the health care system. This has meant fundamental changes in the roles of patients, insurers, providers and the government. Insurers now negotiate with providers on price and quality and patients choose the provider they prefer and join a health insurance policy which best fits their situation. To allow patients to make these choices, much effort has been made to make information on price and quality available to the public. The role of the national government has changed from directly steering the system to safeguarding the proper functioning of the health markets. With the introduction of market mechanisms in the health care sector and the privatization of former sickness funds, the Dutch system presents an innovative and unique variant of a social health insurance system. Since the stepwise realization of the blueprint of the system has not yet been completed, the health care system in the Netherlands should be characterized as being in transition. Many measures have been taken to move from the old to the new system as smoothly as possible. Financial measures intended to prevent sudden budgetary shocks and payment mechanisms have been (and are) continuously adjusted and optimized. Organizational measures aimed at creating room for all players to become accustomed to their new role in the regulated market. As the system is still a "work in progress", it is too early to evaluate the effects and the consequences of the new system in terms of accessibility, affordability, efficiency and quality. Dutch primary care, with gatekeeping GPs at its core, is a strong foundation of the health care system. Gatekeeping GPs are a relatively unusual element in social health insurance systems. The strong position of primary care is considered to prevent unnecessary use of more expensive secondary care, and promote consistency and coordination of individual care. It continues to be a policy priority in the Netherlands. The position of the patient in the Netherlands is strongly anchored in several laws concerning their rights, their relation to providers and insurers, access to information, and possibilities to complain in case of maltreatment. In terms of quality and efficiency of the health care system, the Netherlands is, with some notable exceptions (e.g. implementation of innovations such as day surgery and electronic patient records), an average performer when compared to other wealthy countries. It is too early to tell whether efficiency and quality gains will occur as a result of the 2006 reform.

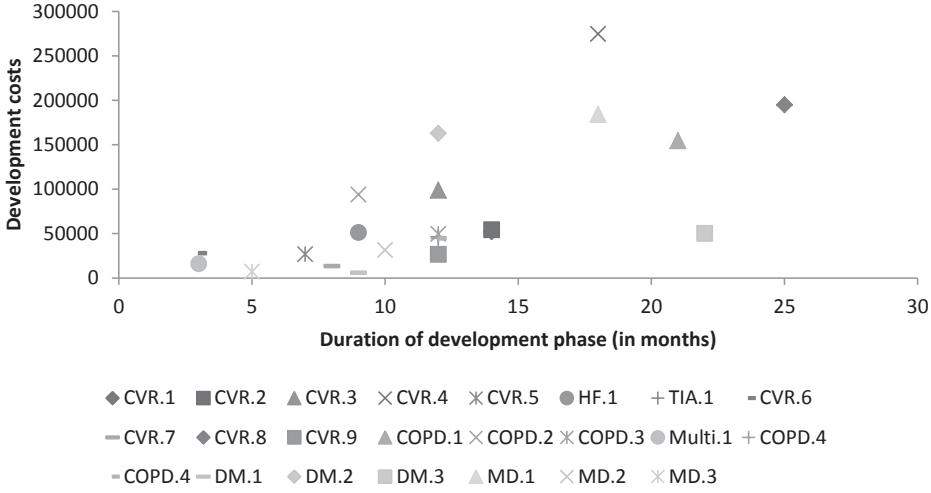
Text from "Schäfer W, Kroneman M, Boerma W, van den Berg M, Westert G, Devillé W and van Ginneken E. The Netherlands: Health system review. Health Systems in Transition, 2010; 12(1):1–229."

Appendix 2 Additional figures

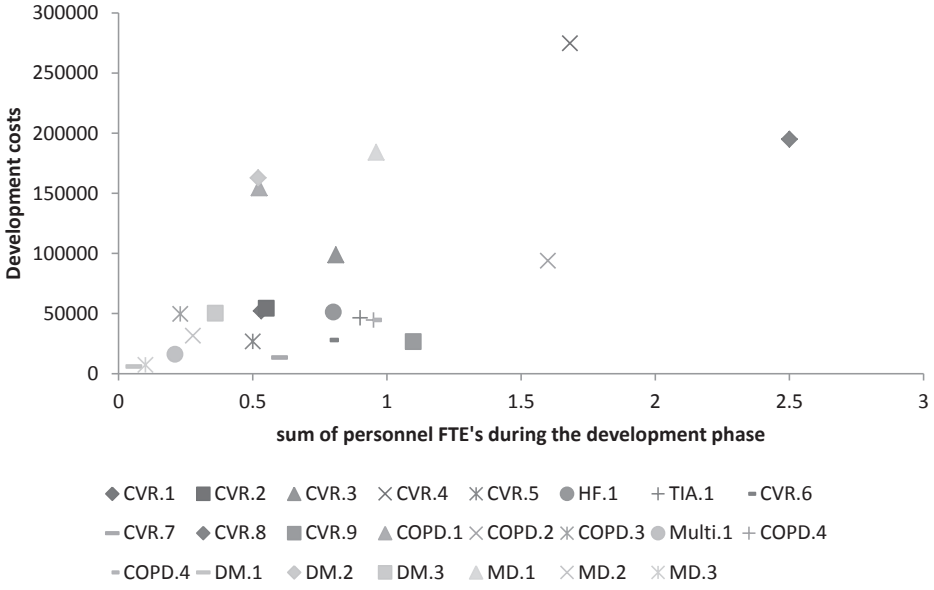
Supplementary Figure 1 This is the relation between development costs and implementation year 1 costs.



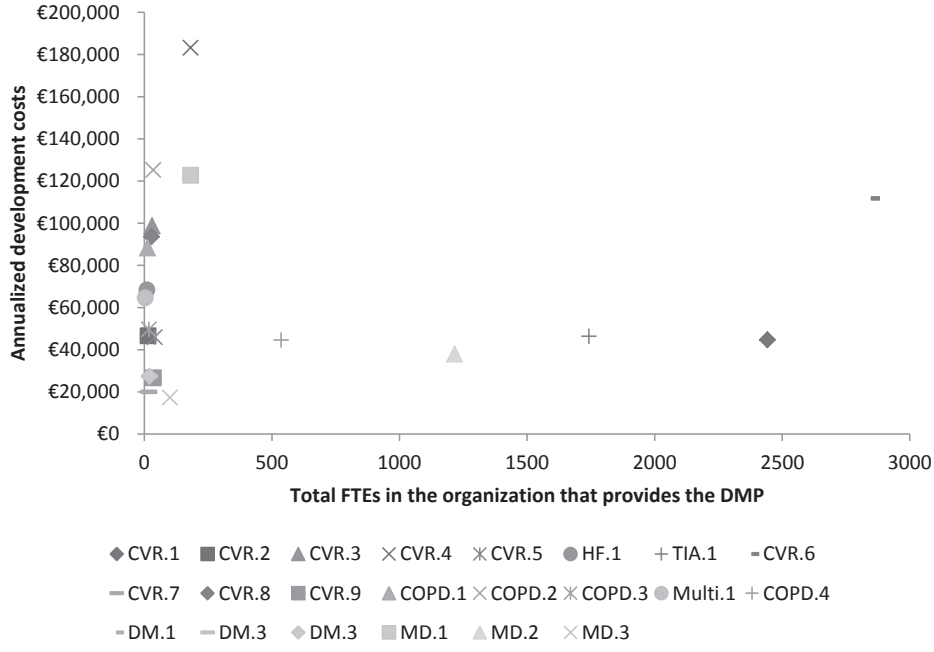
Supplementary Figure 2 Relation between the development costs and the duration of the development phase (in months).



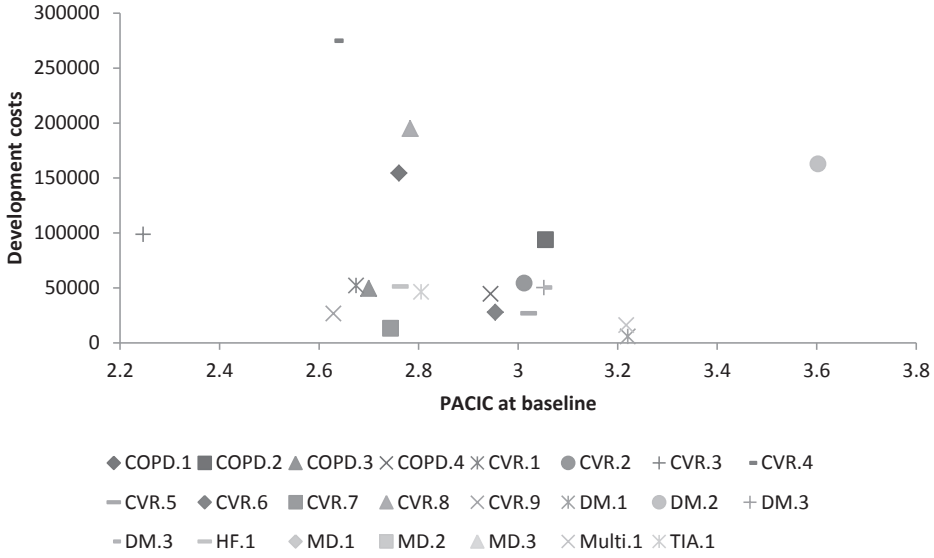
Supplementary Figure 3 Relation between development costs and the number of FTE's dedicated to the development of each DMP



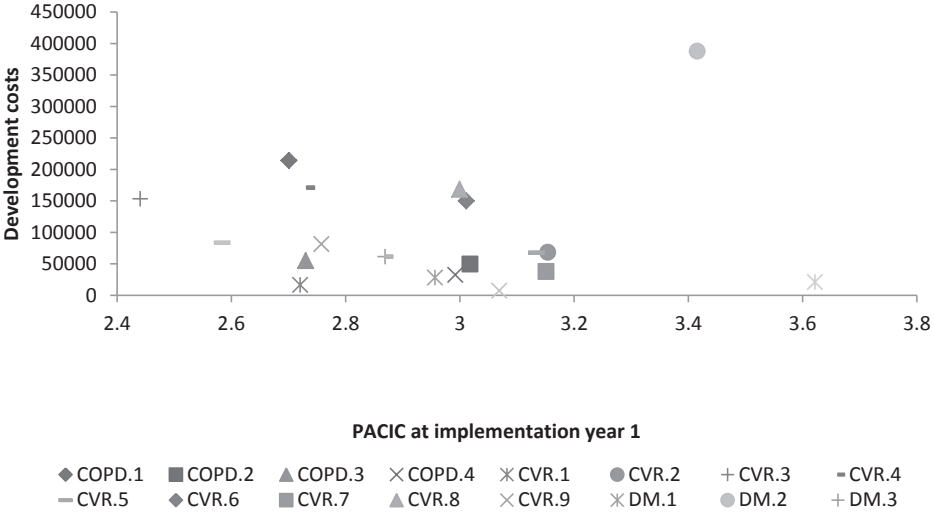
Supplementary Figure 4 Relation between annualized development costs and total number of FTE's in the organization



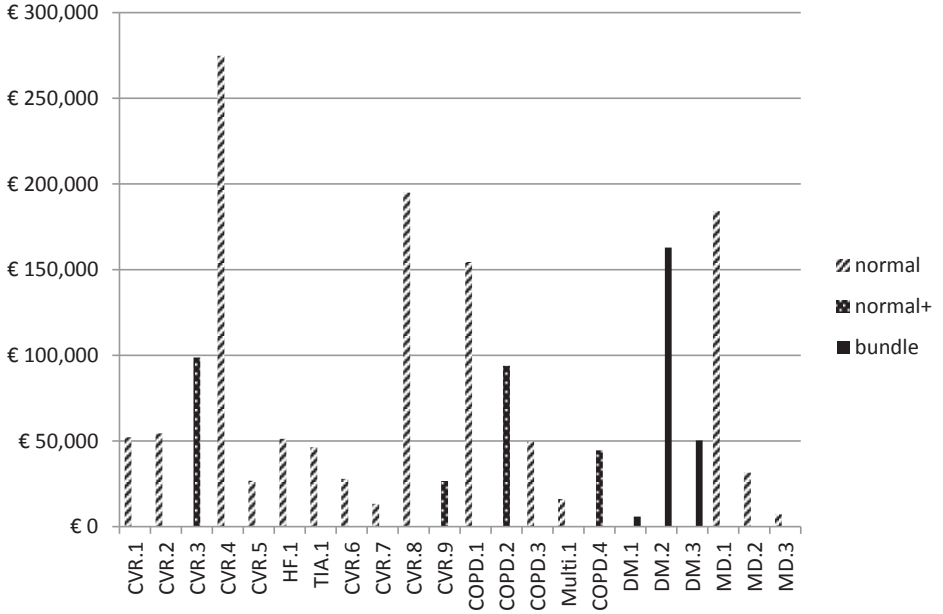
Supplementary Figure 5 Relation between the development costs and PACIC at baseline



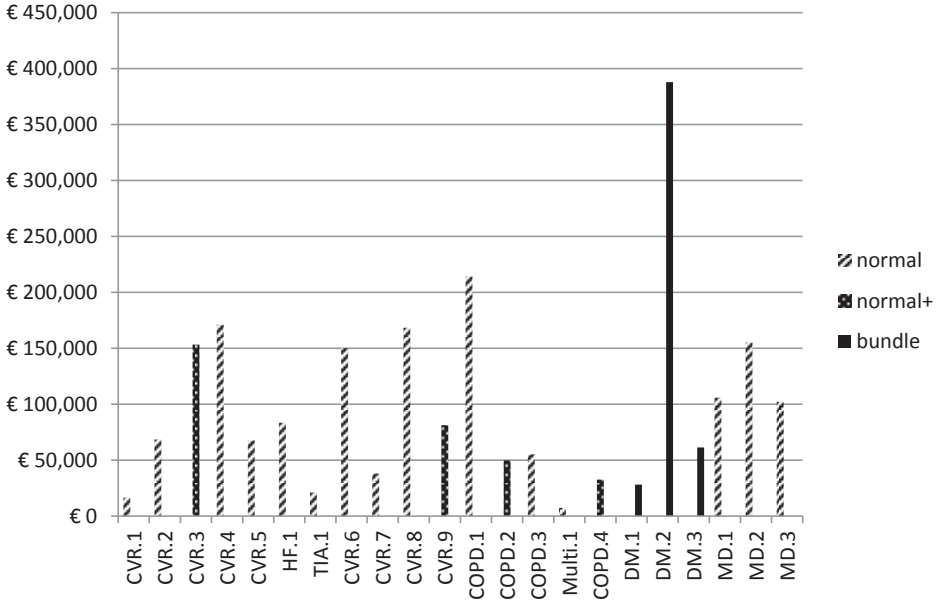
Supplementary Figure 6 Relation between implementation costs and PACIC at year 1



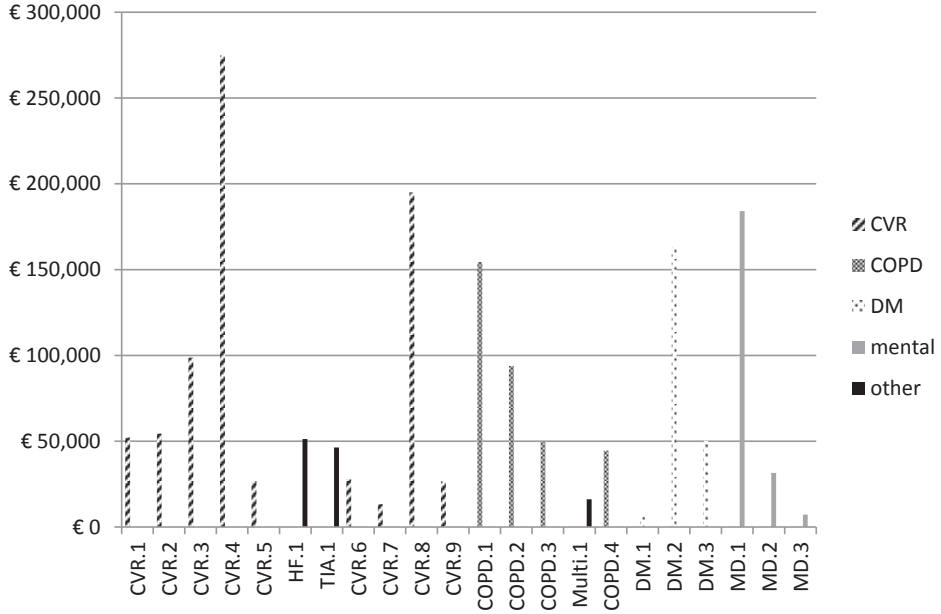
Supplementary Figure 7 Development costs by type of payment at the development phase



Supplementary Figure 8 Implementation costs by payment method at implementation year 1



Supplementary Figure 9 Development costs by disease category



PART C

Economic evaluation of integrated care

CHAPTER 7

Changes in costs and effects after the implementation of disease management programs in the Netherlands: variability and determinants

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Cost Effectiveness and Resource Allocation 2014;12:17

Abstract

Objectives: The aim of the study was to investigate the changes in costs and outcomes after the implementation of various disease management programs (DMPs), to identify their potential determinants, and to compare the costs and outcomes of different DMPs.
Methods: We investigated the 1-year changes in costs and effects of 1,322 patients in 16 DMPs for cardiovascular risk (CVR), chronic obstructive pulmonary disease (COPD), and diabetes mellitus (DMII) in the Netherlands. We also explored the within-DMP predictors of these changes. Finally, a cost-utility analysis was performed from the healthcare and societal perspective comparing the most and the least effective DMP within each disease category.
Results: This study showed wide variation in development and implementation costs between DMPs (range:€16;€1,709) and highlighted the importance of economies of scale. Changes in health care utilization costs were not statistically significant. DMPs were associated with improvements in integration of CVR care (0.10 PACIC units), physical activity (+0.34 week-days) and smoking cessation (8% less smokers) in all diseases. Since an increase in physical activity and in self-efficacy were predictive of an improvement in quality-of-life, DMPs that aim to improve these are more likely to be effective. When comparing the most with the least effective DMP in a disease category, the vast majority of bootstrap replications (range:73%;97%) pointed to cost savings, except for COPD (21%). QALY gains were small (range:0.003;+0.013) and surrounded by great uncertainty.
Conclusions: After one year we have found indications of improvements in level of integrated care for CVR patients and lifestyle indicators for all diseases, but in none of the diseases we have found indications of cost savings due to DMPs. However, it is likely that it takes more time before the improvements in care lead to reductions in complications and hospitalizations.

7.1 Background

Chronic diseases pose an increasing threat to population health, enlarge the burden of care giving, and constrain the financial viability of health care systems worldwide. Because these health care systems originate largely from an era where acute and infectious diseases were more prominent, their design is not optimal for chronic care [23]. This triggered many new approaches for providing continuous, integrated, pro-active and patient-centred care by a multidisciplinary team of care providers in order to improve health outcomes and reduce costs. There is evidence that these approaches improve the quality of the care as measured by process indicators like coordination of care, communication between caregivers, patient satisfaction, provider adherence to guidelines, and patient adherence to treatment recommendations [205]. However, there is debate about the impact on health outcomes and efficiency improvements, a debate complicated by large differences in study designs, outcome metrics and target populations across studies [206] as well as cultural and political barriers to evaluation [207].

In the Netherlands, a recently established regulation introduced a bundled payment system to promote disease management programs (DMPs) for patients with diabetes mellitus type two (DMII), chronic obstructive pulmonary disorder (COPD) or at risk for a cardiovascular disease (CVD) event [47]. Although, the wide-scale implementation of DMII-DMPs was smooth and successful, the uptake of DMPs for COPD and cardiovascular risk (CVR) is still troublesome. This is because health insurers, which contract DMPs from care groups, are yet to be convinced about the financial attractiveness of these programs [93]. Illustrative of this scepticism is that the largest Dutch health insurer does not contract CVR-DMPs and provides only a yearly add-on payment per patient with an elevated CVR to cover costs of coordination, provider training and additional ICT support. Another large health insurer contracts CVR-DMPs only for patients diagnosed with a CVD (secondary prevention) and not for individuals at risk to have CVD (primary prevention). In addition, the debate embeds the adequacy of the current single-disease DMPs for patients with multiple morbidities, which seems to be the norm rather than the exception [208].

Therefore, the provision of evidence about the variability in costs and effects of different implemented DMPs is eminent for the successful implementation of integrated chronic care in the Netherlands. This study aims to investigate the changes in costs and outcomes after the implementation of DMPs, to identify potential determinants of them, and to compare the costs and outcomes of different DMPs.

7.2 Methods

7.2.1 Design and setting

In a prospective pre-post study, we compared 16 different DMPs spread across different regions of the Netherlands [128]: 9 CVR-, 4 COPD-, and 3 DMII-DMPs. Two CVR-DMPs included patients that were at risk for developing CVD (primary prevention), two CVR-DMPs patients that had already been diagnosed with CVD (secondary prevention), and five CVR-DMPs included both patient groups. The implementation of the DMPs and their participation in the evaluation study was financially supported by the Netherlands Organization for Health Research and Development (ZonMw, project number 300030201). Outcomes and health care resource utilization were measured twice, once at the start of the DMP and once after approximately 12 months, using a patient-questionnaire. A detailed description of the design and setting is presented in Lemmens et al., 2011 [128].

7.2.2 Intervention

To describe the details of each DMP we read program documents and interviewed DMP managers using a check-list of possible interventions that may be included in such programs, grouped by the components of the chronic care model [151]. Although the services included in the integrated care package differed between the DMPs, most programs focused on improving the collaboration between different disciplines of health care professionals and redesigning the care-giving process to patient centred care more proactively. Most of them provided interventions such as self-management education and training directed at life-style improvement (physical reactivation, smoking cessation, diet improvement), decision support to implement guidelines and protocols, integration of ICT systems, training for health care providers, case management, and reallocation of tasks between care providers [128,209]. A detailed presentation of the interventions provided by each DMP is provided by Appendix 1.

7.2.3 Outcomes

We investigated the impact of the DMPs on a broad range of outcomes including changes in care delivery process, patient life-style and self-management behaviour, and health-related quality of life (HR-QoL) [151]. More specifically, we investigated the impact of DMPs on: a) the level of chronic care integration using the Patient Assessment Chronic Illness (PACIC) questionnaire [210], b) patient life-style measured by self-reported smoking status (current, former or never smoker) and physical activity (expressed in the number of days per week that an individual had more than 30 minutes physical activity), c) self-efficacy using the respective subscale of the Self-Management Ability Scale-Shorter (SMAS-S) [211], and d) the 3-level EQ-5D utility scores which were based on the Dutch value set and used to estimate quality adjusted life years (QALYs)

[169]. The questionnaire designed to measure these outcomes also included questions about socio-demographic patient characteristics and a checklist of morbidities.

7.2.4 Costs

We estimated five categories of costs, i.e. 1) the development costs, 2) the implementation costs, 3) the costs of health care utilization, 4) the costs borne by patient for travelling to receive care and 5) the costs of productivity loss due to absence from paid work. When calculating costs from a healthcare perspective cost categories 1, 2, and 3 were included; categories 4 and 5 were added when adopting the societal perspective.

The development costs included all costs made during the preparation phase of DMPs e.g. labour costs for brainstorming sessions, training costs, and ICT support costs. The implementation costs were costs that occurred after the provision of DMP interventions to patients had started and included the costs for managing the DMP, the costs of multidisciplinary team meetings, the costs associated with collecting quality of care indicators for audit and feedback, the costs of materials used for patient education, and the costs of keeping the ICT operating. The development and implementation costs were systematically collected using a template based on the CostIt instrument of the World Health Organisation (WHO) [197]. This template was completed during face-to-face interviews with DMPs managers. During these interviews managers were also asked about the presence of additional funding to cover the specific elements of integrated care. Capital costs were amortized over their life span and allocated to the DMP based on square meters for the costs of buildings, full-time equivalents for the costs of ICT and medical technologies (e.g. spirometer). The sum of the capital costs and the operating costs of a DMP was then divided by the number of DMP participants. The costs of developing a DMP were amortized in 5 years assuming this period as the life span of a DMP since after this period changes in guidelines and governmental policies would probably affect the initial form of a DMP. The development and implementation costs per patient were consequently calculated by adding one fifth of the development costs to the annual implementation costs and dividing it by the number of DMP participants.

The costs of health care utilization were based on a questionnaire asking patients about the number of caregiver contacts (GP, nurse practitioner, nurse, dietician, physiotherapist, podiatrist, lifestyle coach, medical specialists in outpatient clinics etc.), hospital admissions and admission days, and medication use. The recall period for these questions was 3 months and we asked for all health care utilization, whether or not it was related to the disease targeted in the DMP. In addition to these costs, the travel costs of patients were calculated, using their self-reported distance to a health care provider. Finally, the costs of productivity loss due to illness were calculated, using the friction cost approach [172], based on questions about absence from paid employment due to

illness. Standard unit costs as reported by [212] were applied. All costs were inflated to 2012 and reported on an annual basis per patient (see Appendix 2).

7.2.5 Statistical analysis to estimate changes within DMPs

We started with paired Wilcoxon tests and McNemar chi-square tests to investigate whether the differences in costs and effects between the baseline and follow-up measurements were statistically significant. In addition, a multi-level analysis was performed to explore the determinants of change in costs and EQ-5D utilities of patients clustered in DMPs. Generalized linear mixed models were used to accommodate the skewness in the health care utilization cost and EQ-5D data as well as to include predictor variables on patient and DMP level. Predictor variables on patient level included: the EQ-5D or costs at baseline (depending which of the two was the outcome variable), age, physical activity at baseline and its change, the PACIC score at baseline and its change, the SMAS-self-efficacy score at baseline and its change, smoking cessation during the follow-up period, and presence of multi-morbidity. Gender, socio-economic status, and marital status were not included in the final model after performing likelihood ratio tests. Predictor variables on the DMP level included the DMP target population and the existence of additional payments to cover overhead and management expenses provided on top of the usual payment per patient.

To explore the variance in the change in outcomes and costs between DMPs that targeted patients at risk for a first (primary prevention), or subsequent CVD event (secondary prevention), or both types of CVR prevention, we also estimated separate models for these sub-groups.

7.2.6 Statistical analysis to estimate differences between DMPs

In each disease category, we identified the DMP that was most effective and least effective in improving the patients' generic health-related quality of life as measured in QALYs. In this manner we identified 5 pairs of DMPs (i.e. for primary CVR prevention, secondary CVR prevention, both types of CVR prevention, COPD, and DMII). For each of the 5 pairs, we calculated the cost-utility of the most effective versus the least effective DMP in terms of incremental costs per QALY gained. These calculations were performed from two perspectives, i.e. the health care perspective (cost category one to three) and the societal perspective (all five categories of costs).

We used inverse probability weighting to balance the two comparators in each pair with respect to age, gender, education, presence of multi-morbidity, marital status, and EQ-5D at baseline. Inverse probability weighting was chosen because it is the preferred propensity score matching technique for small samples [213]. We performed bootstrapping to generate 5,000 samples from the original sample. For each bootstrapped sample we estimated a generalized linear model for each outcome variable (i.e. QALYs or costs)

using the inverse probability weights to get the coefficients adjusted for the propensity score of each observation as well as age, gender, education level, multi-morbidity, and marital status. We used inverse Gaussian distribution and power minus two link for the QALY estimation and gamma distribution and log link for the costs estimation. In this manner, 5,000 predicted incremental costs and 5,000 predicted incremental QALYs were generated. Each of the 5,000 ICERs was calculated as the mean of the predicted incremental costs divided by the mean of the incremental QALYs. These predicted ICERs were then plotted on a cost-effectiveness (CE) plane to show the uncertainty in the ICER.

7.2.7 Sensitivity analysis

The CUA was also performed excluding the development and implementation costs in order to investigate how sensitive the estimated ICERs are to these costs.

7.3 Results

7.3.1 Sample

As Table 1 shows, there were 2,438 respondents at the baseline measurement and 1,974 respondents at the follow-up measurement. One thousand three hundred twenty two individuals responded to both measurements (i.e. had complete data).

The sample characteristics by disease are presented in Table 2. The mean age of the total sample was 65.1 years and consisted of 47% females, 38% low educated, 38% employed, and 30% singles. The mean multi-morbidity among the respondents measured by the Charlson co-morbidity index [170] was 1.83. The COPD sample included proportionally more low-educated, unemployed, and single patients than the other two samples. COPD patients were also older and had higher Charlson co-morbidity scores.

Table 3 presents the baseline values of the outcome measures and their change after one year. The perceived level of chronic care integration was the highest at baseline among patients in DMII-DMPs (3.29) and the lowest in CVR-DMPs (2.80). Individuals in CVR-DMPs were the most physically active at baseline (5.00 days per week) while diabetic patients were the least physically active (4.74 days). In addition, the percentage of smokers was the highest in the COPD sample (39%) and the lowest in the CVR sample

Table 1 Sample size per disease and measurement moment

Disease	DMPs	Baseline	Follow-up	Baseline & follow-up
Total	16	2,438	1,974	1,322
CVR	9	1,342	1,125	725
COPD	4	689	596	395
DMII	3	407	253	202

Table 2 Sample characteristics by disease at baseline

	CVR	COPD	DMII	Total sample
	Mean (sd) [DMP range]	Mean (sd) [DMP range]	Mean (sd) [DMP range]	Mean (sd) [DMP range]
Age	64.1 (9.7) [59.6; 67.8]	66.5 (10.0) [65.4;69.3]	66.2 (9.7) [64.2;67.1]	65.1** (9.9) [59.6;69.3]
% Females	48	48	43	47
Charlson comorbidity index	1.48 (1.10)	2.26 (1.28)	2.22 (0.99)	1.83** (1.20) [1.15;2.48]
% Low education	35	48	25	37**
% Employment	43	30	37	38**
% Single	26	36	30	30**

The table presents the mean (sd) unless otherwise indicated; in [ ] is given the range between DMPs i.e. lowest and highest values across DMPs in the same disease area; low education was defined as no or only primary education; The p-values show whether the values are statistically different between the diseases ** Statistically different at p< 0.01 between the diseases.

(21%). Patients in DMII-DMPs had scored the highest in self-efficacy (4.56) and patients in COPD-DMPs the lowest (4.33). The mean EQ-5D utility score at baseline was 0.83 in the CVR sample and 0.84 in the DMII sample while for the COPD sample it was lower (0.79).

7.3.2 Changes in outcomes

Changes in PACIC scores were significantly positive (0.10) in the CVR sample (range across the 9 CVR DMPs from +0.02 to +0.26) and significantly negative (-0.23) in the DMII sample (range across the 3 DMII-DMPs from -0.27 to -0.18). In the CVR and COPD samples the change in the number of days per week with more than 30 minutes of physical activity was positive and statistically significant (0.33 and 0.37 respectively). The range in physical active days across the CVR and COPD-DMPs was quite large as Table 3 shows. The percentage of smokers decreased substantially in all samples (ranging across all 16 DMPs from -13.7 percentage points to -2.5 percentage points) as well as the self-efficacy (ranging from -0.48 percentage points to 0.15 percentage points) and the HR-QoL (ranging from -0.06 percentage points to +0.03 percentage points).

7.3.3 Changes in costs

The development and first year's implementation costs per patient of the 16 DMPs are presented in Table 4. As this table shows, there is large variation in the implementation costs per patient between and within the three diseases ranging from €16 to €1,709. This is due to the variation in the total development and implementation costs and the number of participants per DMP. The largest share of these costs is for costs related to the time that personnel dedicates to the implementation of DMPs. Costs related to educational courses for caregivers and information brochures for patients were low in

Table 3 Outcomes by disease at baseline and differences with the outcomes in the follow-up

	CVR			COPD			DMII			Total sample	
	Mean at baseline (sd)	Mean change (sd)	Range of change across DMPs[#]	Mean at baseline (sd)	Mean change (sd)	Range of change across DMPs[#]	Mean at baseline (sd)	Mean change (sd)	Range of change across DMPs[#]	Mean change	Range of change across DMPs[#]
PACIC (1; 5 highest=best)	2.80 (0.84)	0.10** (0.80)	+0.02; +0.26	2.92 (0.89)	-0.03 (0.75)	-0.05; +0.06	3.29 (0.85)	-0.23** (0.72)	-0.27; -0.18	0.01 (0.78)	-0.27; +0.26
Physically active days per week	5.00 (2.07)	0.33** (2.15)	-0.23; +0.82	4.82 (2.13)	0.37** (2.20)	-0.11; +1.36	4.74 (1.94)	0.29 (2.01)	+0.05; +0.89	0.34** (2.14)	-0.23; +1.36
% smokers	21	-6pp**	-2.5pp; -10.7pp	39	-11pp**	-7.3pp;-13.7pp	22	-9pp**	-8pp; -13.6pp	-8pp**	-13.7pp; -2.5pp
Self-efficacy (1; 6 highest=best)	4.45 (0.87)	-0.28** (0.75)	-0.33; -0.15	4.33 (0.88)	-0.34** (0.73)	-0.48; -0.27	4.56 (0.85)	-0.29** (0.77)	-0.42; -0.22	-0.30** (0.75)	-0.48; -0.15
EQ-5D (-0.33; 1 highest=best)	0.83 (0.18)	-0.01* (0.16)	-0.06; +0.03	0.79 (0.20)	-0.04** (0.19)	-0.04; –0.03	0.84 (0.16)	-0.03* (0.14)	-0.04; -0.02	-0.02** (0.17)	-0.06; +0.03

pp= percentage points; * ($p< 0.05$); ** ($p<0.01$); the differences are calculated subtracting the outcome values at baseline from the outcome values at follow-up

Table 4 Development and implementation costs by DMP

	N	Development phase*			Total costs without amortization #	Implementation Year 1* Costs per patient without amortization	Costs per patient with amortization
		Total costs without amortization #	Costs per patient without amortization	Costs per patient with amortization*			
CVR-DMP 1	300	52,136	174	35	16,426	55	90
CVR-DMP 2	207	54,417	263	53	68,415	331	381
CVR-DMP 3	700	98,754	141	28	153,215	219	234
CVR-DMP 4	300	274,783	916	183	171,026	570	605
CVR-DMP 5	550	26,807	49	10	67,604	123	142
CVR-DMP 6	450	27,923	62	12	149,990	333	356
CVR-DMP 7	125	13,324	107	21	37,968	304	387
CVR-DMP 8	250	195,007	780	156	168,385	674	715
CVR-DMP 9	1,000	26,678	27	5	81,258	81	92
COPD-DMP 1	2,508	154,504	62	12	214,239	85	90
COPD-DMP 2	1,600	93,909	59	12	49,751	31	38
COPD-DMP 3	133	49,639	373	75	55,191	415	493
COPD-DMP 4	2,400	44,586	19	4	32,599	14	18
DMII-DMP 1	2,400	5,891	2	0	28,061	12	16
DMII-DMP 2	233	162,889	699	140	387,879	1,655	1,709
DMII-DMP 3	300	50,304	168	34	61,338	204	239

*We used 5 years as amortization period; # These costs are not per patient

almost all cases (except in DMII-DMP1). In some DMPs "other" costs such as ICT, energy, and accommodation costs were relatively high (e.g. 66% in DMII-DMP 2).

At baseline, patients in COPD-DMPs had the highest mean yearly hospital costs (€1,967), medication costs (€857), total health care costs (€4,368) and total costs (€5,320) while patients in CVR-DMPs had the highest mean yearly productivity loss (€1,648) (see Table 5). Patients in DMII-DMPs had the highest primary care costs (€941). However, almost all differences between baseline and follow-up were statistically insignificant and the standard deviations of the estimated means were large. Only the outpatient costs of patients with diabetes increased by €115. As Table 5 shows, the changes across DMPs within the same disease and between diseases varied largely. The cost change within each disease category ranged from negative to positive across DMPs except for the outpatient costs and inpatient costs of patients with diabetes.

In primary and mixed prevention CVR-DMPs, the PACIC was increased by 0.18 and 0.10 and the number of days with at least 30 minutes of physical activity in a week increased by 0.43 and 0.37, respectively (Table 6). The decrease in the percentage of smokers ranged from 3% (primary prevention) to 8% (secondary prevention). As Table

Table 5 Costs at baseline and differences with the follow-up measurement

	CVR			COPD			DMII			Total sample	
	Mean at baseline (sd)	Mean change (sd)	Range of change across DMPs	Mean at baseline (sd)	Mean change (sd)	Range of change across DMPs	Mean at baseline (sd)	Mean change (sd)	Range of change across DMPs	Mean change	Range of change across DMPs
Primary care	610 (857)	34 (1,069)	-510; +314	916 (1388)	49 (1,601)	-5; +155	941 (947)	-84 (1,226)	-236; +88	21 (1,273)	-510; +314
Outpatient hospital care	365 (778)	30 (954)	-443; +259	654 (2,488)	-119 (2,524)	-272; +22	338 (604)	115* (809)	+86; +169	-2* (1,583)	-443; +259
Inpatient hospital care[$]	587 (3,526)	624 (9,452)	-551; +2,148	1,967 (13,256)	320 (18,563)	-396; +1,162	701 (3,714)	-454 (4,065)	-1,211; -220	368 (12,426)	-1,211; +2,148
Medication	370 (362)	3 (261)	-45; +41	857 (601)	3 (417)	-2; +6	518 (482)	1 (318)	-44; +34	3 (323)	-45; +41
Total healthcare utilization costs	1,911 (4,102)	691 (9,812)	-1,107; +2,626	4,368 (14,256)	238 (19,080)	-672; +1,055	2,504 (4,015)	-446 (4,444)	-93; -1,066	382 (12,826)	-1,107; +2,626
Travelling	74 (215)	-2 (344)	-113; +90	226 (1,190)	-109 (1,145)	-328; +47	174 (378)	-22 (441)	-23; -19	-37** (699)	-328; +90
Productivity	1,648 (8,080)	-495 (7,349)	-1,988; +1,075	658 (4,724)	341 (6,603)	0; +459	216 (1,410)	188 (2,656)	-210; +454	-102 (6,571)	-1,988; +1,075
Total costs	3,302 (9,006)	468 (13,559)	-1,893; +4,269	5,320 (15,390)	85 (20,354)	-1,232; +375	3,489 (7,605)	-517 (9,662)	-1,591; -167	203 (15,448)	-1,893; +4,269

$ inpatient hospital care costs include also emergency care costs; * ($p< 0.05$); ** ($p<0.01$); the differences are calculated subtracting the costs at baseline from the costs at follow-up; primary care costs included contacts with GP, nurse practitioner, nurse, dietician, physiotherapist, podiatrist, lifestyle coach, etc

Table 6 Costs and outcomes by type of CVR prevention

	Primary prevention		Secondary prevention		Mixed	
	Baseline	Change	Baseline	Change	Baseline	Change
PACIC (1-5 highest)	2.64 (0.77)	0.18* (0.76)	2.52 (0.79)	0.09 (0.75)	2.92 (0.84)	0.10* (0.82)
Physically active days per week	5.25 (1.91)	0.43* (1.94)	5.15 (2.10)	0.12 (2.11)	4.91 (2.10)	0.37** (2.20)
% smokers	13	-3*	30	-8**	20	-6**
Self-efficacy (1-6 highest)	4.44 (0.85)	-0.29** (0.75)	4.32 (0.92)	-0.30** (0.77)	4.48 (0.86)	-0.27** (0.74)
EQ-5D	0.85 (0.17)	-0.01 (0.15)	0.77 (0.22)	0.01 (0.19)	0.84 (0.17)	-0.02* (0.15)
Primary care costs	555 (827)	-16 (701)	810 (1,153)	-149 (1,191)	565 (751)	97 (1,092)
Outpatient hospital care	326 (662)	-104 (643)	725 (1,342)	-34 (1,728)	269 (492)	76* (657)
Inpatient hospital care[$]	471 (3,009)	-334 (3,120)	1,064 (5,012)	932 (9,807)	476 (3,085)	742 (10,225)
Medication costs	269 (275)	0 (248)	493 (423)	1 (289)	356 (351)	4 (255)
Total healthcare utilization costs	1,600 (3,665)	-447 (3,663)	3052 (5,787)	754 (10,204)	1,653 (3,525)	918 (10,574)
Travelling costs	63 (145)	73 (571)	89 (221)	-48* (185)	72 (226)	-5* (312)
Productivity costs	3,542 (11,480)	-1,685 (10,076)	1,119 (6,401)	-86 (6,964)	1,405 (7,646)	-368 (6,743)
Total costs	3,633 (10,091)	-317 (11,593)	4,421 (10,657)	159 (13,876)	2,911 (8,201)	725 (13,874)

The table presents the mean (SD) and the mean difference (SD) between baseline and follow-up measurements; $ inpatient hospital care costs include also emergency care costs; * ($p< 0.05$); ** ($p<0.01$); the differences are calculated subtracting the costs at baseline from the costs at follow-up; primary care costs included contacts with GP, nurse practitioner, nurse, dietician, physiotherapist, podiatrist, lifestyle coach, etc

6 shows, self-efficacy was decreased in all three types of CVR prevention by about 0.28 while the EQ-5D decreased in the mixed CVR prevention DMPs by 0.02.

Table 6 presents the yearly costs and outcomes of patients enrolled in CVR-DMPs that target different populations (i.e. primary prevention, secondary prevention, or both types of prevention). After 12 months, the hospital costs of patients included in DMPs targeting both types of CVR prevention increased by €819 within a year. Further, patients in DMPs for secondary prevention and for both types of prevention had €48 and €5 lower travelling costs, respectively. The travelling costs at baseline in these two types of DMPs were also higher compared to the primary prevention DMPs.

7.3.4 Determinants of changes in HR-QoL and costs within DMPs

The results from the generalized linear mixed models are presented in Table 7. Model one shows that a greater improvement in EQ-5D utility is significantly predicted by a lower baseline EQ-5D score, a higher baseline physical activity level, a greater increase in physical activity, and a greater increase in self-efficacy. One additional day with more than 30 minutes of physical activity leads to a 3% higher EQ-5D utility and 1 unit of in-

Table 7 Determinants of changes in HR-QoL and health care utilization costs

	Model 1 Change in EQ-5D		Model 2 Change in health care utilization costs	
	e(b)	p	e(b)	p
Intercept	1.04	0.744	104192.98	<0.001
EQ-5D/	0.60	<0.001		
Costs (in 000's) baseline			0.95	<0.001
Age	1.00	0.408	1.00	0.130
Physical activity (1-7 highest)	1.02	0.023	1.00	0.777
Change in physical activity	1.03	0.001	1.00	0.639
PACIC (1-5 highest)	0.99	0.474	1.02	0.247
Change in PACIC	1.00	0.830	1.00	0.843
Self-efficacy (1-6 highest)	1.00	0.956	0.98	0.107
Change self-efficacy	1.04	0.032	1.01	0.730
Quit smoking (1= yes)	1.04	0.119	1.07	0.104
Multi-morbidity (1=yes)	0.95	0.019	1.06	<0.001
COPD* (1=yes)	0.93	<0.001	1.01	0.541
DMII* (1=yes)	0.99	0.576	1.02	0.460
Additional payment (1=yes)	0.99	0.468	0.99	0.491
N	820		843	
R^2 patient level	0.36		0.73	
R^2 DMP level	0.56		0.78	

* the reference category is CVR-DMP; Note: the predictor variables COPD-DMP, DMII-DMP, and Additional payment are on the DMP level. All other variables are on the patient level.

crease in self-efficacy score leads to a 4% higher EQ-5D utility. In contrast, patients with COPD had 7% less improvement in EQ-5D and patients with multi-morbidity 5% less.

The best predictors of change in health care utilization costs were health care utilization costs at baseline and the presence of multi-morbidity (model 2). If costs were €1000 higher at baseline, the increase was 5% less. In case of multi-morbidity, the cost increase was 6% higher. The variance in the dependent variables explained by models 1 and 2 at the DMP and the patient level was relatively high.

7.3.5 Comparing costs and effects between DMPs

The results from the cost-utility analysis taking the health care and societal perspective are presented in Table 8. This table shows that the most effective DMP for CVR primary prevention, combined primary and secondary CVR prevention, and DMII led to statistically significant cost savings when compared to the least effective DMP in the same disease category (i.e. more than 95% of bootstrap replications in the southern quadrants). It also shows there is large variation in incremental costs (ranging from €-721 to

Table 8 Results from the cost-utility analysis

	Most effective VS least effective DMP *	Incremental costs	Incremental QALYs	Mean ICER	% of 5000 simulated ICERs per quadrant in the CE plane			
					NW	NE	SW	SE
Health care perspective								
CVR-primary[#]	7 VS 4	-534 (297)	0.003 (0.021)	-178,539	1	3	41	56
CVR-secondary[$]	1 VS 3	-671 (976)	0.012 (0.015)	-56,809	6	21	15	58
CVR-both	2 VS 8	-721 (416)	0.005 (0.016)	-148,480	2	2	35	61
COPD	1 VS 4	1,716 (2,000)	0.009 (0.053)	185,747	33	46	11	10
DMII	1 VS 3	-677 (398)	0.013 (0.013)	-50,234	1	3	14	82
Societal perspective								
CVR-primary[#]	7 VS 4	-1,131 (1,334)	0.003 (0.021)	-377,991	5	12	37	46
CVR-secondary[$]	1 VS 3	-153 (1,225)	0.012 (0.015)	-12,929	10	36	11	43
CVR-both	2 VS 8	-604 (554)	0.005 (0.016)	-124,457	6	8	31	55
COPD	1 VS 4	2,054 (2,371)	0.009 (0.053)	-222,314	34	47	11	9
DMII	1 VS 3	-1,735 (1,084)	0.013 (0.013)	-128,790	1	2	14	83

* most effective is defined based on the highest incremental QALY and the reverse; [#] primary prevention for CVD; [$]secondary prevention for CVD; ICER: incremental cost-effectiveness ratio; CE: cost-effective(ness); best is defined as most effective based on QALYs and worse as the least effective based on the same measurement; the numbers correspond to the DMP numbers in Table 4.

€1,716) and incremental QALYs (ranging from 0.003 to 0.013) between the best and the worst DMP within a disease category. Due to the very small incremental QALYs the ICERs are very large. The 5000 bootstrapped ICERs plotted on the CE plane showed that there is large uncertainty around the estimated mean ICER. Considering the CVR-primary prevention sample, 97% of the 5,000 simulated ICERs were in the southern half of the CE plane indicating lower incremental costs while the reverse was observed for the COPD sample (79% of the 5,000 bootstrapped ICERS were on the Northern CE plane).

From the societal perspective, the cost-utility results are similar to the results from the health care perspective except that for the secondary CVR prevention samples the uncertainty about the incremental costs became even larger.

7.3.6 Sensitivity analysis

Table 9 shows the results from the CUA performed excluding the development and implementation costs. The most remarkable change in comparison to the main CUA is that 20% (instead of 4%) of the 5,000 bootstrapped ICERs regarding both CVR prevention DMPs were located on the North quadrant of the CE plane. This change is a result from the higher development and implementation costs of the least effective DMP.

Table 9 Results from the cost-utility analysis from the health care perspective excluding the development and implementation costs

	Best DMP VS worse DMP*	Incremental costs	Incremental QALYs	Mean ICER	% of 5000 simulated ICERs per quadrant in the CE plane			
					NW	NE	SW	SE
CVR-primary[#]	7 VS 4	-407 (330)	0.003 (0.021)	-136,077	3	7	39	51
CVR-secondary[$]	1 VS 3	-863 (961)	0.012 (0.015)	-73,013	4	14	17	65
CVR-both	2 VS 8	-326 (388)	0.005 (0.016)	-67,145	10	10	28	52
COPD	1 VS 4	1,574 (1,985)	0.009 (0.053)	170,390	32	45	12	11
DMII	1 VS 3	-430 (402)	0.013 (0.013)	-31,942	3	11	12	74

* most effective is defined based on the highest incremental QALY and the reverse; [#] primary prevention for CVD; [$]secondary prevention for CVD; ICER: incremental cost-effectiveness ratio; CE: cost-effective(ness); best is defined as most effective based on QALYs and worse as the least effective based on the same measurement; the numbers correspond to the DMP numbers in Table 4.

7.4 Discussion

In this study we have investigated the short-term changes in costs and effects after the implementation of 16 DMPs for three different chronic diseases, namely CVR, COPD, and DMII. We have also explored the within DMP predictors of these changes. Finally, a CUA was performed from the health care and societal perspective comparing each DMP to usual care and comparing the most effective and least effective DMP within five disease categories (i.e. CVR-primary prevention, CVR-secondary prevention, CVR-both types of prevention, COPD, DMII).

Our results show a significant improvement in the level of chronic care integration as measured by the PACIC, in the CVR population (0.10). It improved especially in the DMPs that were directed at primary prevention (0.18) or the combination of primary and secondary prevention (0.10) of cardiovascular diseases. This is promising because patients in these programs had the lowest PACIC scores of the three patient groups. For

patients who already had a cardiovascular disease it is probably harder to achieve improvements in integrating care because more (para-) medical disciplines and healthcare sectors become involved. An unexpected result was that the PACIC decreased by 0.23 in the DMII-DMPs. This may be due to difficulties to maintain their high starting level of integrated care, which in turn may be caused by the attention that was paid to quality improvements in diabetes care for the last decade. It would be interesting to examine whether our findings would have been similar if another instrument, for example the Assessment of Chronic Illness Care (ACIC), would have been used to measure the level of chronic care integration. However, we did not include the ACIC in our analysis for two reasons. The first is because this paper focuses on intermediate and final outcomes in patients, not in professionals. The second is that although the two instruments are complementary [214], they both measure the level of integrated care and thus, they correlate [215].

Another interesting finding is that DMPs seem to improve the life-style of patients, in all three disease categories. Patients reported a higher level of physical activity, especially those in DMPs for COPD and CVR management. In addition, the percentage of smokers decreased by more than 5 percentage-point in all disease categories; the decrease was 11 percentage-point in COPD. This reduction is considerably higher as the cessation rate achieved by a physician-advice to stop smoking [216] or the impact of the recent ban on smoking in bars and restaurants [217].

Furthermore, our within-DMP analysis showed a reduction in self-efficacy and generic HR-QoL after the implementation of the DMPs. The slight deterioration (about 0.03 EQ-5D units) in HR-QoL may be explained as a time effect rather than a treatment effect because the HR-QoL of chronic care patients generally tends to decrease over time [218]. Similarly, the decrease in self-efficacy may also be related to the decrease of HR-QoL because deterioration in HR-QoL may worsen self-efficacy [219,220]. Another explanation may be that HR-QoL and self-efficacy are both perceived values that are influenced by the information and knowledge a patient has. DMP interventions included educating patients about their disease, learning them to recognize the early signals of disease-worsening, learning them coping skills and stimulating them to improve their lifestyle. As a result, patients may have become more aware of their impaired health status and their reference point may have shifted.

Our study collected the costs of development and implementation of the DMPs in detail and showed that they can be an important driver of total costs. This is in line with the findings of the few previous studies that have incorporated them in their analysis [27,153,206]. The development and implementation costs per patient were largely driven by the personnel costs. Moreover, the 16 DMPs included in our sample were pioneers in experimenting with DMPs. Therefore, the number of enrolled patients was perhaps not as high in the first year of implementation as the capacity would allow. In the long(er)

term, we expect that more patients will be enrolled in the DMPs and caregivers will gain experience in managing and maintaining a DMP. That may lower the implementation costs per patient. Therefore, we would expect more favourable ICERs for the DMPs in the longer term. Within the one-year time frame of our study there are as yet few signals of important changes in the costs of healthcare utilization and productivity loss. But the heterogeneity in DMPs is large with all 3 DMII-DMPs showing a numerical reduction of hospital costs and total health care costs.

The regression analysis indicated that an increase in physical activity was predictive of an increase in HR-QoL. Given the observed increase in physical activity in almost all disease categories, we may expect DMPs to improve HR-QoL in the longer term. We also found that an improvement in self-efficacy was predictive of an improvement in HR-QoL. This creates an opportunity for DMPs to develop and implement strategies to improve the self-efficacy of the patients. Furthermore, patients with multiple morbidities seem to benefit less than patients with one disease. This may imply that the current disease-specific DMPs do not address the needs that patients with multi-morbidity have, and therefore, are less effective for this population. The need for patient-tailored care to address the complex needs of patients with multi-morbidity is extensively addressed in the literature [221,222]. A horizontal integration of DMPs to simultaneously target CVR, COPD, and DMII might be appealing for several reasons. The first one is of course the desire to improve the care for these patients. The second reason is that some components of the DMPs are largely similar, irrespective of the disease. For example, smoking cessation support and physical reactivation can be organized similarly, and adjusted to the specific needs of an individual patient. This avoids inefficiencies and double payments. Another reason is that the number of participants in such a multi-disease DMPs will increase, which will lower the implementation and overhead costs per participant.

We also performed a CUA comparing DMPs within a disease area, which is interesting for decision makers once they have decided to implement a DMP. Then the variability in costs and health outcomes is likely to drive the choice of program. When adopting the health care perspective the CUA showed that the majority of the bootstrapped ICERs in all types of CVR prevention and DMII comparison pairs were located on the South-East quadrant of the CE plane. This indicates that the most effective DMPs had lower costs and positive QALY gains compared to the least effective DMPs in these three disease groups. This finding remained also when the societal perspective was adopted. However, the results concerning the primary CVR prevention and COPD were more difficult to interpret because of the uncertainty about the QALY gains (health care and societal perspective).

As our results showed, the cost-effectiveness of DMPs varies considerably, most likely depending on the components of the program, the target population, the success of the implementation and the costs of managing and operating the program. These are

all factors that contractors of DMPs should consider in the negotiation phase. We are planning future analysis aiming to identify the factors that drive the cost-effectiveness of a DMP. These findings could contribute to the on-going debate in the Netherlands on whether the current bundled payment system for single-disease DMPs are an intermediate stage towards population-based financing [93]. Population based financing includes a risk-adjusted fixed budget (either per group of patients or region) to cover all health care provided by multiple professionals from different disciplines. Savings compared to a pre-defined benchmark are often shared between payer and provider. A large variation in the cost-effectiveness of DMPs due to the aforementioned factors, jeopardizes the successful implementation of DMPs as means to achieve integration of chronic care. Thus, a population-based financing with larger scope in terms of covered population and provided interventions, economies of scale that lower operating costs, and consensus of all stakeholders that ensures successful implementation may appear attractive to Dutch policy makers. However, the preconditions to introduce a population-based financing are far from being reached [223] and therefore, the implementation of DMPs on more disease areas is still work in progress.

This study contributes to the growing body of international evidence on integrated care in several ways. First, it highlights the necessity to adopt a broad set of outcome measures and include the most important cost items from different perspectives in the evaluation of DMPs. Second, the findings of our study support the previous studies that concluded that DMPs are positively associated with improvements in patient lifestyle and quality of care [215,224,225]. Third, our finding that DMPs have the potential to become cost-effective in the long-term, and the identification of factors that drive that cost-effectiveness, could inform designers of integrated care programs in other European countries. Fourth, the limitation of disease-specific DMPs to address the needs of complex patients could urge collective initiatives on a European level to develop adequate models of integrated care for this population.

Our study is one of very few studies providing insight into health economic aspects of DMPs that includes such a broad range of outcome measures and cost categories. However, we fully acknowledge the limitations of the study design with respect to causality. At the start of this study there were multiple initiatives to provide integrated care across the entire country, stimulated by the introduction of the bundled payment system and other financial incentives. Therefore it was impossible to create a control group at regional level. It was also difficult to identify control groups within the same organization because of the high risk of contamination [152]. This risk is high because the implementation of a DMP requires changes at an organisational level. For example, redesigning the care-delivery process or training nurses in motivational interviewing affects the entire organisation and the entire target population. Therefore, we did not aim to compare the DMPs to usual care but rather compare different DMPs within a

disease category. To optimize comparability, we applied inverse probability weighting and corrected for confounders in multivariate analysis. In addition, our results may be object to regression to the mean bias. However, this bias is probably limited because our sample size is relatively large and the diseases included in our analysis are chronic and progressive. These assumptions are supported by a previous study that found minimal evidence of regression to the mean in COPD-DMPs [226].

7.5 Conclusions

This study of the short-term effects of DMPs found that the implementation of DMPs was associated with improvements in integration of care and lifestyle behaviour, such as physical activity and smoking, of patients with CVR, diabetes and COPD. Since an increase in physical activity and an increase in self-efficacy were predictive of an improvement in HR-QoL, DMPs that aim to improve these are more likely to be effective. This study has also shown the wide variation in development and implementation costs between DMPs and pointed at the importance of economies of scale. On this short term we have not found statistically significant cost savings due to DMPs, but it is likely that it takes more time before the improvements in care lead to reductions in complications and hospitalizations.

Appendix 1 Unit cost prices used in the costs analysis

	2009	2010	2011	2012
Inflation rate - Dutch Statistics (CBS)	1,2	1,3	2,3	2,5
GP session	28	28	29	30
GP home visit	43	44	45	46
GP phone contact	14	14	15	15
Inpatient day	457	463	474	485
Day care treatment	251	254	260	267
Intensive care unit day	2,183	2,211	2,262	2,319
Outpatient visit	72	73	75	76
Emergency room visit	151	153	156	160
Physical therapy	36	36	37	38
Speech therapy (session)	33	33	34	35
Occupational therapy (hour)	22	22	23	23
Dietary advice (hour)	27	27	28	29
Home care (hour)	35	35	36	37
cost/km	0.20	0.20	0.21	0.21

CHAPTER 8

Cost-effectiveness of disease management programs for cardiovascular risk and COPD in the Netherlands

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submitted for review

Abstract

OBJECTIVES: Disease management programs (DMPs) for cardiovascular risk (CVR) and chronic obstructive pulmonary disease (COPD) are increasingly implemented in the Netherlands to improve care and patient's health behaviour. The aim of the study was to provide evidence about the (cost-)effectiveness of Dutch DMPs as implemented in daily practice.

METHODS: We compared the physical activity, smoking status, quality-adjusted life years (QALYs) and yearly costs per patient between the most and the least comprehensive DMP in four disease categories: primary CVR-prevention, secondary CVR-prevention, both types of CVR-prevention, and COPD (n:1,034). Propensity score matching increased comparability between DMPs. A two-year cost-utility analysis (CUA) was performed from the healthcare and societal perspective. Sensitivity analysis was performed to estimate the impact of DMP development and implementation costs on the cost-effectiveness.

RESULTS: Patients in the most comprehensive DMPs increased their physical activity more (except for the primary CVR-prevention) and had higher smoking cessation rates. The incremental QALYs ranged from -0.032 to 0.038 across all diseases. From a societal perspective, the most comprehensive DMP decreased costs in the primary CVR-prevention (certainty: 57%), secondary CVR-prevention (certainty: 88%), and both types of CVR-prevention (certainty: 98%). Moreover, the implementation of comprehensive DMPs lead to QALY gains in secondary CVR-prevention (certainty: 92%) and COPD (certainty: 69%).

CONCLUSIONS: The most comprehensive DMPs for CVR and COPD have the potential to be cost-saving, effective, or cost-effective compared to the least comprehensive DMPs. The challenge for Dutch stakeholders is to find the optimal mixture of interventions that is most suited for each target group.

8.1 Introduction

In the Netherlands, a bundled payment scheme was introduced in 2010 to promote disease management programs (DMPs) for patients with diabetes mellitus, chronic obstructive pulmonary disorder (COPD) or at risk for a cardiovascular disease event (CVR) [47]. The aim of this new scheme was to improve quality and efficiency in chronic care by providing continuous, integrated, pro-active, patient-centred, and comprehensive care. DMPs were seen as the means to achieve such improvements by including interventions that target the organisation of health care delivery, health care professionals, and patients [227].

The wide-scale implementation of diabetes DMPs was smooth and successful. However, the uptake of DMPs for COPD and CVR is more troublesome. One of the reasons is that health insurers, which contract DMPs from care groups are not convinced about the financial attractiveness of these programs [93]. The wide variability in the number and type of interventions across the implemented DMPs [228] is another reason of scepticism. Previous studies found that DMPs with more interventions are more likely to save costs in health care [37,39]. However, there is no clear distinction between DMPs in the Netherlands based on their comprehensiveness.

Illustrative of the reluctance among Dutch health care insurers is that the largest insurer does not contract CVR-DMPs and provides only a yearly add-on payment per patient with an elevated CVR to cover costs of coordination, provider training and additional ICT support. Another large health insurer contracts CVR-DMPs only for patients diagnosed with a cardiovascular disease (CVD) (secondary prevention) and not for individuals at risk to have CVD (primary prevention).

Previous studies found there is evidence that CVR-DMPs and COPD-DMPs improve process indicators like coordination of care and communication between caregivers [205,225]. Whether this translates into better health outcomes for patients and less costs for the healthcare system needs to be investigated. This study aims to provide empirical evidence about the comparative (cost-) effectiveness of the most versus least comprehensive DMPs that have been implemented in a particular disease area. This evidence is required for the successful implementation of integrated chronic care in the Netherlands.

8.2 Methods

8.2.1 Design and setting

In a prospective two-year observational study, we compared 13 different DMPs provided by primary care groups spread across different regions of the Netherlands [128]: 9 CVR-

and 4 COPD-DMPs. Two CVR-DMPs included patients that were at risk for developing CVD (primary prevention), two CVR-DMPs patients that had already been diagnosed with CVD (secondary prevention), and five CVR-DMPs included both patient groups. The COPD-DMPs included patients with mild to moderate COPD severity (i.e. Global Initiative for chronic Obstructive Lund Disease (GOLD) stage 1 and 2). The implementation of the DMPs and their participation in the evaluation study was financially supported by the Netherlands Organization for Health Research and Development (ZonMw, project number 300030201). Outcomes and health care resource utilization were measured at baseline (2010), 12 months (2011), and 24 (2012) months using a patient-questionnaire. A detailed description of the design and setting is presented in Lemmens et al., 2011 [128].

8.2.2 Intervention

All DMPs were developed based on Wagner's Chronic Care Model (CCM) [25,229]. To describe the details of each DMP we studied program documents and interviewed DMP managers using a check-list of possible interventions that may be included in such programs; these interventions were grouped by the components of the CCM [151]. Most programs focused on improving the collaboration between different disciplines of health care professionals and redesigning the care-giving process towards pro-active, patient-centred care. Most of them provided interventions such as self-management education and training directed at improving healthy behaviour (physical reactivation, smoking cessation, diet improvement, treatment adherence), decision support to implement guidelines and protocols, audit and feedback, integration of ICT systems, training for health care providers, case management, and reallocation of tasks between care providers [128,209,230,231]. However, the number of interventions included in the integrated care package differed considerably between the DMPs. We rank ordered all DMPs in the same disease area according to the number of different interventions included. The one with the highest number was defined as the most comprehensive and the one with the lowest number as the least comprehensive. This way we created 4 pairs of DMPs (i.e. for primary CVR-prevention, secondary CVR-prevention, both types of CVR-prevention, and COPD) that were compared in the analyses.

8.2.3 Outcomes

We investigated the impact of the DMPs on patients' healthy behaviour, measured by self-reported smoking status (current, former or never smoker) and physical activity (number of days in a week that a patient was physically active for more than 30 minutes). These outcomes were selected because healthy behaviour and self-management were two of the primary aims of all DMPs, either to lower the CVR or to reduce disease progression in COPD. We also measured the 3-level EQ-5D utility scores which were based

on the Dutch value set to estimate quality adjusted life years (QALYs) [169]. QALYs were calculated as the area under the EQ-5D utility curve over time. The questionnaire designed to measure these outcomes also included questions about socio-demographic patient characteristics and a checklist of morbidities.

8.2.4 Costs

We estimated five categories of costs, i.e. 1) the costs of health care utilization, 2) the costs borne by patient for travelling to receive care and 3) the costs of productivity loss due to absence from paid work, 4) the development costs of the DMPs, and 5) the implementation costs of the DMPs.

The costs of health care utilization were based on a questionnaire asking patients about the number of caregiver contacts (GP, nurse practitioner, nurse, dietician, physiotherapist, podiatrist, lifestyle coach, medical specialists in outpatient clinics etc.), hospital admissions and admission days, and medication use. The recall period for these questions was 3 months and we asked for all health care utilization, whether or not it was related to the disease targeted in the DMP. In addition to these costs, the travel costs of patients were calculated, using their self-reported distance to a health care provider. Finally, the costs of productivity loss due to illness were calculated, using the friction cost approach [172], based on questions about absence from paid employment due to illness. The duration of the friction period was 115 days. Standard unit costs as reported by [212] were applied (see Appendix 1). All costs were inflated to 2012 and reported on an annual basis per patient.

The development costs included the costs that were made during the preparation phase of DMPs e.g. labour costs for brainstorming sessions, training costs, and ICT support costs. The implementation costs were costs that occurred after the provision of DMP interventions to patients had started and included the costs for managing the DMP, the costs of multidisciplinary team meetings, the costs associated with collecting quality of care indicators for audit and feedback, the costs of materials used for patient education, and the costs of keeping the ICT operating. The development and implementation costs were systematically collected using a template based on the CostIt instrument of the World Health Organisation (WHO) [197]. This template was completed during face-to-face interviews with DMPs managers. During these interviews managers were also asked about the presence of additional funding to cover the specific elements of integrated care. Capital costs were amortized over their life span and allocated to the DMP based on square meters for the costs of buildings, and full-time equivalents for the costs of ICT and medical equipment (e.g. spirometer). The sum of the capital costs and the operating costs of a DMP was then divided by the number of patients participating in the DMP. The costs of developing a DMP were amortized in 5 years, recognizing that optimizing a DMP is an ongoing process. The combined development and implementation costs

per patient were consequently calculated by adding one fifth of the development costs to the annual implementation costs and dividing it by the number of DMP participants.

8.2.5 Propensity score matching

As a consequence of the observational study design, the patients in the comparators in each disease category differed in disease severity (as expressed in quality of life) and socio-demographic characteristics at baseline. Therefore, we used propensity score matching to reduce confounding caused by these differences. Inverse probability weighting was used to balance the most comprehensive and least comprehensive DMP in each pair with respect to age, gender (Male/Female), low education (Yes/No), presence of multi-morbidity (Yes/No), single as marital status (Yes/No), and EQ-5D at baseline. Inverse probability weighting was chosen because it is the preferred propensity score matching technique for small samples [213]. Stabilized inverse propensity scores were used as weights in order to preserve the sample size of the original data and to produce appropriate estimations of variance [232].

8.2.6 Estimated differences and cost-utility analysis (CUA)

For each of the four pairs, we performed bootstrapping to generate 5,000 samples from the original sample. For each bootstrap sample, repeated measurement analysis was performed by using Generalized Estimating Equations (GEE) models to estimate the difference in mean level of physical activity, proportion of current smokers, QALYs and yearly costs per patient from the health care perspective (including cost category 1) and the societal perspective (including cost categories 1, 2, and 3) at baseline, 12 months, and 24 months for each pair of comparators. The mean predicted values were adjusted for age, gender, education level, presence of multi-morbidity, EQ-5D and marital status at baseline as well as the stabilised inverse probability weight. We used a Gaussian distribution and identity link for the physical activity and QALY estimation, binomial distribution and logit link for the smoking estimation, and a gamma distribution and a log link for the costs estimation.

Based on the predicted outcomes, we calculated the cost-utility of the most comprehensive versus the least comprehensive DMP in terms of incremental costs per QALY gained in each disease sample. In this manner, 5,000 predicted incremental costs and 5,000 predicted incremental QALYs were generated over a two year period (i.e. costs and QALYs at the first and second year were summed). The point estimate of the incremental cost-effectiveness ratio (ICER) was calculated as the mean of the predicted incremental costs divided by the mean of the predicted incremental QALYs. The predicted incremental costs and predicted incremental QALYs were then plotted on a cost-effectiveness (CE) plane to show the uncertainty in the ICER.

8.2.7 Sensitivity analysis

The CUA was also performed including the development and implementation costs per patient (cost categories 4 and 5) in order to investigate how sensitive the estimated ICERs are to these costs. When the implementation costs in year 2 were not available (i.e. in two DMPs), we linearly extrapolated the costs from implementation year 1.

8.3 Results

8.3.1 Defining the DMP comprehensiveness

The number of interventions per component of the CCM for each DMP is presented in Table 1. As this table shows, decision support, delivery system design and self-management were the CCM components with the most interventions, followed by organizational support, ICT and community. Regarding primary CVR-prevention, CVR-DMP 4 was the most comprehensive (43 interventions) and CVR-DMP 7 was the least comprehensive (23 interventions). CVR-DMP 3 (29 interventions) was the most comprehensive and CVR-DMP 1 (23 interventions) was the least comprehensive in secondary CVR-prevention. Among the DMPs for both types of CVR-prevention, CVR-DMP 6 was the most comprehensive (35 interventions) and CVR-DMP 9 was the least comprehensive (13 interventions). Moreover, in COPD, COPD-DMP 4 was the most

Table 1 Number of interventions per DMP

	ORG	COM	SM	DS	DSD	ICT	Total	Comprehensive*
CVR-DMP 1 (secondary CVR-prevention)	0	4	6	6	6	1	23	Least
CVR-DMP 2 (both types of CVR-prevention)	0	3	5	6	5	5	24	
CVR-DMP 3 (secondary CVR-prevention)	1	7	3	6	7	5	29	Most
CVR-DMP 4 (primary CVR-prevention)	3	7	6	12	9	6	43	Most
CVR-DMP 5 (both types of CVR-prevention)	1	4	6	7	5	6	29	
CVR-DMP 6 (both types of CVR-prevention)	2	7	6	6	8	6	35	Most
CVR-DMP 7 (primary CVR-prevention)	2	1	6	6	7	1	23	Least
CVR-DMP 8 (both types of CVR-prevention)	0	5	6	5	4	5	25	
CVR-DMP 9 (both types of CVR-prevention)	0	0	3	4	3	3	13	Least
COPD-DMP 1	2	4	2	8	6	6	28	Least
COPD-DMP 2	2	6	6	7	6	3	30	
COPD-DMP 3	1	7	7	7	6	3	31	
COPD-DMP 4	3	6	8	11	7	4	39	Most
Total	17	61	70	91	79	54		

* within a disease category; ORG: organizational support; COM: community; SM: self-management; DS: decision support; DSD: delivery system design; ICT: information and communication technology; CVR: cardiovascular risk; COPD: chronic obstructive pulmonary disease; DMP: disease management program

comprehensive (39 interventions) and COPD-DMP 1 was the least comprehensive (28 interventions).

8.3.2 Sample characteristics

The sample size, patient characteristics at baseline and baseline values of the outcome measures of the most and the least comprehensive DMP within a disease category are presented in Table 2. Most patient characteristics differed between the DMPs in each disease category. Compared to the CVR DMPs, the COPD DMPs included patients with a higher co-morbidity score as measured by the Charlson co-morbidity index [170]. The COPD DMPs also included more low-educated, unemployed, currently smoking patients

Table 2 Sample size, patient characteristics and predicted outcome measurements at baseline after propensity score matching

	Primary CVR-prevention		Secondary CVR-prevention		Both types of CVR-prevention		COPD	
	Most C. CVR-DMP 4	Least C. CVR-DMP 7	Most C. CVR-DMP 3	Least C. CVR-DMP 1	Most C. CVR-DMP 6	Least C. CVR-DMP 9	Most C. COPD-DMP 4	Least C. COPD-DMP 1
N baseline	133	34	112	120	257	157	88	133
N 12 months	84	20	58	80	161	90	61	93
N 24 months	64	20	33	58	147	104	51	93
Mean age (sd)	63.1 (10.5)	60.1 (11.3)	65.2 (8.0)	67.6 (10.5)	63.2 (10.4)	66.3 (10.3)	66.5 (9.9)	65.1 (11.4)
% Females	57	56	36	43	54	42	43	45
Mean Charlson index (sd)	1.4 (1.0)	1.6 (1.1)	1.9 (1.2)	1.7 (1.2)	1.6 (1.2)	1.4 (1.3)	2.1 (1.3)	2.4 (1.3)
% low education	41	44	38	40	44	31	45	56
% employment	45	59	34	35	46	42	35	27
% single	26	32	35	46	21	26	34	34
Mean physical activity (sd) [#]	4.78 (0.17)	5.60 (0.36)	4.89 (0.18)	4.84 (0.22)	4.37 (0.14)	4.57 (0.18)	4.56 (0.24)	4.92 (0.17)
% smokers[#]	0.132	0.234	0.280	0.326	0.163	0.185	0.352	0.452
QALY[#]	0.823 (0.016)	0.836 (0.020)	0.753 (0.015)	0.751 (0.015)	0.820 (0.010)	0.820 (0.010)	0.761 (0.015)	0.759 (0.015)
Mean costs: health care (sd) [#]	1,769 (339)	1,732 (491)	3,377 (559)	2,982 (545)	3,130 (789)	2,840 (622)	4,928 (1,130)	6,194 (1,351)
Mean costs: societal (sd) [#]	4,520 (2,370)	6,444 (3,376)	4,847 (882)	4,430 (1,100)	4,939 (1,060)	3,707 (771)	6,820 (1,505)	7,757 (1,692)

Notes: Most C. and Least C. refer to most comprehensive and least comprehensive respectively; CVR: cardiovascular risk; COPD: Chronic Obstructive Pulmonary; This table reports the patient characteristics at baseline; Low education was defined as no or only primary education; [#] predicted values after propensity score marching

and costly patients than the CVR DMPs. Among the CVR DMPs, patients in the secondary CVR-prevention DMPs had the highest mean age, mean co-morbidity score, and mean costs as well as proportionally more low educated, males, employed, single, currently smoking patients. Patients in the secondary CVR-prevention DMPs also had the lowest mean QALY score at baseline among all DMPs.

8.3.3 Healthy behaviour

The results showed that the predicted physical activity increased over time in all DMPs. The level of physical activity was higher in the most comprehensive DMPs compared to the least comprehensive DMPs in three disease categories (Panel A of Table 3). Only patients in the least comprehensive primary CVR-prevention DMP were on average 0.15 days at the first year and 0.34 days at the second year more physically active than the patients in the most comprehensive primary prevention-CVR DMP. However, in all cases the differences between the comparators were not statistically significant. Panel B of Table 3 shows that the proportion of current smokers decreased over time in all DMPs except for CVR-DMP 9. It also shows that there were proportionally less current smokers in the most comprehensive DMPs at the first year (range across diseases samples: 2%-7%) and the second year (range across diseases samples: 1%-7%) than in the least comprehensive DMPs. These differences were not statistically significant.

Table 3 Predicted healthy behaviour after propensity score matching

Panel A: Predicted physical activity	First year Mean [CI]	Second year Mean [CI]
Primary CVR-prevention (n:305;c:151)		
CVR-DMP 4 (most comprehensive)	5.31 [4.94;5.69]	5.76 [5.37;6.16]
CVR-DMP 7 (least comprehensive)	5.46 [4.58;6.19]	6.10 [5.33;6.81]
Difference	-0.15 [-0.98;0.79]	-0.34 [-1.15;0.54]
Secondary CVR-prevention (n:365;c:200)		
CVR-DMP 3 (most comprehensive)	5.14 [4.70;5.57]	5.23 [4.59;5.83]
CVR-DMP 1 (least comprehensive)	4.89 [4.44;5.31]	5.25 [4.69;5.81]
Difference	0.26 [-0.34;0.86]	-0.02 [-0.83;0.79]
Both types of CVR-prevention (n:737;c:359)		
CVR-DMP 6 (most comprehensive)	5.11 [4.81;5.41]	5.52 [5.23;5.80]
CVR-DMP 9 (least comprehensive)	5.00 [4.59;5.39]	5.47 [5.12;5.82]
Difference	0.11 [-0.38;0.61]	0.05 [-0.39;0.50]
COPD (n:470;c:198)		
COPD-DMP 4 (most comprehensive)	5.31 [4.88;5.72]	5.33 [4.88;5.74]
COPD-DMP 1 (least comprehensive)	4.92 [4.58;5.26]	5.13 [4.67;5.59]
Difference	0.40 [-0.13;0.91]	0.19 [-0.45;0.83]

Table 3 Predicted healthy behaviour after propensity score matching (continued)

Panel B: Predicted proportion of current smokers	First year % smoking [CI]	Second year % smoking [CI]
Primary CVR-prevention (n:332;c:155)		
CVR-DMP 4 (most comprehensive)	0.099 [0.046;0.153]	0.069 [0.024;0.114]
CVR-DMP 7 (least comprehensive)	0.166 [0.052;0.301]	0.135 [0.030;0.267]
Difference	-0.067 [-0.211;0.057]	-0.066 [-0.212;0.046]
Secondary CVR-prevention (n:425;c:212)		
CVR-DMP 3 (most comprehensive)	0.205 [0.129;0.279]	0.215 [0.125;0.304]
CVR-DMP 1 (least comprehensive)	0.259 [0.183;0.334]	0.276 [0.185;0.370]
Difference	-0.055 [-0.158;0.051]	-0.061 [-0.188;0.063]
Both types of CVR-prevention (n:854;c:387)		
CVR-DMP 6 (most comprehensive)	0.133 [0.092;0.173]	0.119 [0.078;0.159]
CVR-DMP 9 (least comprehensive)	0.153 [0.089;0.217]	0.191 [0.123;0.260]
Difference	-0.020 [-0.100;0.055]	-0.072 [-0.151;0.005]
COPD (n:484;c:210)		
COPD-DMP 4 (most comprehensive)	0.303 [0.212;0.393]	0.292 [0.202;0.379]
COPD-DMP 1 (least comprehensive)	0.328 [0.241;0.416]	0.302 [0.218;0.387]
Difference	-0.025 [-0.147;0.098]	-0.010 [-0.124;0.108]

Notes: n: number of observations; c: number of patients; CI: confidence interval; CVR: cardiovascular risk; COPD: Chronic Obstructive Pulmonary; The difference is calculated by subtracting the values of the least comprehensive DMP from the values of the most comprehensive DMP.

8.3.4 QALYs

The predicted mean QALYs are presented in Table 4. In most cases, the QALYs decreased over time except for CVR-DMP 7 where the predicted mean QALY increased from 0.829 in the first year to 0.832 in the second year. The predicted mean QALYs were higher in the most comprehensive compared to the least comprehensive DMP in the secondary CVR-prevention (by 0.019) in the first and the second year after DMP implementation. In COPD, the most comprehensive DMP had higher (by 0.016) predicted mean QALYs in the first implementation year than the least comprehensive COPD-DMP. However, this difference disappeared in the second year. The difference in the predicted QALYs between the comparators in the both types of CVR-prevention, favoured equally (by 0.017) the least comprehensive DMP in the first implementation year and the most comprehensive DMP in the second year. The least comprehensive DMP in primary CVR-prevention had higher predicted mean QALYs in the first year (by 0.010) and in the second year (by 0.022) than its comparator. In all diseases the differences were not statistically significant.

Table 4 Predicted QALYs after propensity score matching

	First year Mean [CI]	Second year Mean [CI]
Primary CVR-prevention (n:300;c:155)		
CVR-DMP 4 (most comprehensive)	0.820 [0.791;0.848]	0.809 [0.773;0.846]
CVR-DMP 7 (least comprehensive)	0.829 [0.786;0.868]	0.832 [0.768;0.907]
Difference	-0.010 [-0.045;0.029]	-0.022 [-0.102;0.044]
Secondary CVR-prevention (n:402;c:212)		
CVR-DMP 3 (most comprehensive)	0.761 [0.732;0.789]	0.737 [0.700;0.774]
CVR-DMP 1 (least comprehensive)	0.742 [0.710;0.772]	0.719 [0.671;0.764]
Difference	0.019 [-0.006;0.043]	0.019 [-0.030;0.068]
Both types of CVR-prevention (n:756;c:384)		
CVR-DMP 6 (most comprehensive)	0.793 [0.770;0.815]	0.777 [0.749;0.806]
CVR-DMP 9 (least comprehensive)	0.810 [0.0.786;0.833]	0.760 [0.709;0.804]
Difference	-0.017 [-0.038;0.004]	0.017 [-0.029;0.071]
COPD (n:454;c:210)		
COPD-DMP 4 (most comprehensive)	0.752 [0.719;0.783]	0.721 [0.679;0.762]
COPD-DMP 1 (least comprehensive)	0.736 [0.703;0.769]	0.723 [0.682;0.764]
Difference	0.016 [-0.014;0.046]	-0.002 [-0.049;0.043]

Notes: n: number of observations; c: number of patients; CI: confidence interval; CVR: cardiovascular risk; COPD: Chronic Obstructive Pulmonary; The difference is calculated by subtracting the values of the least comprehensive DMP from the values of the most comprehensive DMP.

8.3.5 Costs from the health care and societal perspective

The mean predicted yearly costs per patient are presented in Table 5. For primary CVR-prevention, the difference in healthcare costs was €-1,045 (favouring the most comprehensive DMP) in the first year and €425 in the second year. For secondary CVR-prevention, the difference in healthcare cost favoured the most comprehensive DMP in the first year (€-2,340) and in the second year (€-66). Similarly, the differences in healthcare costs favoured the most comprehensive DMP for both types of CVR-prevention (€-720 in the first year and €-3,221 in the second year). In COPD, healthcare costs in the most comprehensive DMP were higher (by €1,787) in the first year and slightly lower (by €24) in the second year than in the least comprehensive DMP. The results from the societal perspective were similar, except for primary CVR-prevention were the most comprehensive DMP had lower costs per patient (by €1,045) in the first year of implementation compared to the least comprehensive DMP. The most comprehensive COPD-DMP had higher costs in the second year compared to the least comprehensive DMP from the societal perspective. The difference in costs from the health care perspective in the first year in primary CVR was the only statistically significant difference.

Table 5 Predicted yearly costs per patient after propensity score matching

	Health care perspective		Societal perspective	
	First year Mean [CI]	Second year Mean [CI]	First year Mean [CI]	Second year Mean [CI]
Primary CVR-prevention (n:341;c:155)				
CVR-DMP 4 (most comprehensive)	1,398 [978;1,971]	1,344 [879;1,978]	3,051 [1,306;6,609]	2,034 [827;4,786]
CVR-DMP 7 (least comprehensive)	1,161 [619;1,886]	920 [501;1,541]	4,096 [759;10,682]	1,683 [468;5,092]
Difference	237 [-477;937]	425 [-228;1,702]	-1,045 [-7,413;3,421]	351 [-1,665;2,032]
Secondary CVR-prevention (n:440;c:214)				
CVR-DMP 3 (most comprehensive)	2,354 [1,501;3,234]	3,225 [1,238;6,940]	3,201 [1,955;4,582]	4,031 [1,488;8,780]
CVR-DMP 1 (least comprehensive)	4,693 [2,599;7,419]	3,291 [1,694;5,434]	5,543 [3,058;8,904]	5,156 [2,477;8,501]
Difference	-2,340 [-5,138;-64]	-66 [-3,148;4,117]	-2,342 [-5,834;566]	-1,125 [-5,594;4,438]
Both types of CVR-prevention (n:885,c:389)				
CVR-DMP 6 (most comprehensive)	3,386 [1,791;6,183]	2,509 [1,563;4,085]	4,228 [2,454;6,793]	2,741 [1,867;3,995]
CVR-DMP 9 (least comprehensive)	4,106 [2,585;6,469]	5,730 [1,946;12,156]	5,568 [3,325;8,533]	8,007 [2,715;19,385]
Difference	-720 [-2,663;1,461]	-3,221 [-9,509;394]	-1,341 [-4,096;1,262]	-5,266 [-16,464;51]
COPD (n:502;c:211)				
COPD-DMP 4 (most comprehensive)	6,688 [3,321;11,614]	3,281 [2,397;4,374]	8,355 [4,097;14,168]	3,738 [2,680;5,101]
COPD-DMP 1 (least comprehensive)	4,901 [3,002;6,858]	3,305 [2,530;4,209]	5,498 [3,691;7,624]	3,553 [2,693;4,577]
Difference	1,787 [-1,794;6,404]	-24 [-1,185;1,199]	2,857 [-1,582;8,508]	185 [-1,264;1,787]

Notes: n: number of observations; c: number of patients; CI: confidence interval; CVR: cardiovascular risk; COPD: Chronic Obstructive Pulmonary; The difference is calculated by subtracting the values of the least comprehensive DMP from the values of the most comprehensive DMP.

8.3.6 Development and implementation costs

As Table 6 shows, the total costs and per patient costs of the DMP development and implementation were higher in the most comprehensive DMPs compared to the least comprehensive DMPs except for the COPD category.

8.3.7 Results from the cost-utility analysis

The results from the CUA are presented in Table 7. The incremental QALYs ranged from -0.032 in primary CVR-prevention to 0.038 in secondary CVR-prevention. Taking

Table 6 Development and implementation costs by disease management programme

	Number of eligible patients	Development phase*			Implementation Year 1*			Implementation Year 2*		
		Total costs without amortization#	Costs per patient without amortization	Costs per patient with amortization*	Total costs without amortization#	Costs per patient without amortization	Costs per patient with amortization	Total costs without amortization#	Costs per patient without amortization	Costs per patient with amortization
Primary CVR-prevention										
CVR-DMP 4 (most comp.)	300	274,783	916	183	171,026	570	605	176,068	587	622
CVR-DMP 7 (least comp.)	125	13,324	107	21	37,968	304	387	26,328	211	294
Secondary CVR-prevention										
CVR-DMP 3 (most comp.)	700	98,754	141	28	153,215	219	234	112,686	161	176
CVR-DMP 1 (least comp.)	300	52,136	174	35	16,426	55	90	-	-	-
Both types CVR-prevention										
CVR-DMP 6 (most comp.)	450	27,923	62	12	149,990	333	356	122,432	272	295
CVR-DMP 9 (least comp.)	1,000	26,678	27	5	81,258	81	92	58,441	58	69
COPD										
COPD-DMP 4 (most comp.)	2,400	44,586	19	4	32,599	14	18	24,464	10	15
COPD-DMP 1 (least comp.)	2,508	154,504	62	12	214,239	85	90	-	-	-

*We used 5 years as amortization period; # These costs are not per patient; CVR: cardiovascular risk; COPD: Chronic Obstructive Pulmonary; comp.: comprehensive;

the health care perspective (i.e. Panel A of Table 7), the most comprehensive DMP for CVR-primary prevention led to higher costs (certainty: 87%) when compared to the least comprehensive DMP. In contrast, the most comprehensive DMP providing secondary CVR-prevention and both types of CVR-prevention led to less costs (certainty: 78% and 96%, respectively). Moreover, the vast majority of the 5,000 simulated ICERs were located in the Eastern quadrant of the CE plane in the secondary CVR-prevention (91%) and the COPD (69%) denoting QALY gains from the implementation of comprehensive DMPs. When QALY gains were very small, the ICERs became very high.

Table 7 Results from the two-year CUA

	Most comprehensive versus least comprehensive DMP	Incremental Costs (€)	Incremental QALYs	Mean ICER (€)	% of 5000 simulated ICERs per quadrant in the CE plane			
					NW	NE	SW	SE
Panel A: Health care perspective								
CVR-prim[#]	CVR-DMP 4 VS CVR-DMP 7	661 (621)	-0.032 (0.045)	-20,515	67	20	9	3
CVR-sec[$]	CVR-DMP 3 VS CVR-DMP 1	-2,405 (2,347)	0.038 (0.028)	-63,980	1	13	8	78
CVR-both	CVR-DMP 6 VS CVR-DMP 9	-3,940 (2,684)	<0.001 (0.026)	-11,571,968	2	2	49	47
COPD	COPD-DMP 4 VS COPD-DMP 2	1,763 (2,287)	0.014 (0.027)	127,659	23	54	8	15
Panel B: Societal perspective								
CVR-prim[#]	CVR-DMP 4 VS CVR-DMP 7	-694 (2,988)	-0.032 (0.045)	21,526	28	14	48	9
CVR-sec[$]	CVR-DMP 3 VS CVR-DMP 1	-3,467 (3,115)	0.038 (0.028)	-92,220	1	12	8	80
CVR-both	CVR-DMP 6 VS CVR-DMP 9	-6,607 (4,443)	<0.001 (0.026)	-19,402,211	1	1	50	48
COPD	COPD-DMP 4 VS COPD-DMP 1	3,042 (2,929)	0.014 (0.027)	220,338	26	59	5	10
Panel C: Sensitivity analysis: health care perspective including development and implementation costs								
CVR-prim[#]	CVR-DMP 4 VS CVR-DMP 7	1,203 (603)	-0.032 (0.045)	-37,320	75	22	2	1
CVR-sec[$]	CVR-DMP 3 VS CVR-DMP 1	-2,011 (2,336)	0.038 (0.028)	-53,486	1	17	7	75
CVR-both	CVR-DMP 6 VS CVR-DMP 9	-2,806 (2,609)	<0.001 (0.026)	-8,240,387	6	6	45	43
COPD	COPD-DMP 4 VS COPD-DMP 1	1,527 (2,262)	0.014 (0.027)	110,579	22	52	9	17

[#] primary CVR-prevention; [$]secondary CVR-prevention; CVR = cardiovascular risk; COPD= Chronic Obstructive Pulmonary Disease; HC = Health Care perspective; SP = Societal Perspective; NE = North East; NW = North West; SW = South West; SE = South East. ICER: incremental cost-effectiveness ratio; CE: cost-effectiveness; in brackets are standard deviations presented;

As Panel B of Table 7 shows, the results from the CUA taking the societal perspective were similar to the results from the health care perspective except for the primary CVR-prevention. In this sample, 57% of the simulated ICERs indicated that the most comprehensive DMP led to cost savings instead of higher costs as found in the CUA from the health care perspective.

The CE planes are presented in Appendix 2 and the observed yearly costs and QALYs are presented in Appendix 3.

8.3.8 Sensitivity analysis

The results from the CUA taking the health care perspective and including the development and implementation costs are presented in Panel C of Table 7. These results showed that the addition of development and implementation costs reduced the cost savings of the most comprehensive program in secondary and both types of CVR-prevention. Inclusion of these costs did not lead to noteworthy differences in the allocation of the 5,000 simulated ICERs on the quadrants of the CE plane.

8.4 Discussion

In this study we have investigated the differences in physical activity, smoking status, QALYs and costs between the most and the least comprehensive DMP in four disease categories: primary CVR-prevention, secondary CVR-prevention, both types of CVR-prevention, and COPD. A CUA was performed from the health care and societal perspective. Sensitivity analysis was performed to estimate the impact of DMP development and implementation costs on the cost-effectiveness.

Our study is one of very few studies providing evidence about the cost-effectiveness of DMPs for COPD and CVR in the Netherlands. We purposely did not aim to compare DMPs to usual care, because at the start of this study there were so many initiatives to provide integrated care across the entire country that it was impossible to construct a usual care group. Moreover, the decision to implement DMPs had already been taken and was stimulated by the nationwide introduction of the bundled payment system and other financial incentives [47]. Therefore, many DMP interventions have been diffused in usual care during the follow-up period. This is supported by the results of a cluster RCT that evaluated a COPD-DMP compared with usual care, were the authors concluded that usual care had moved into the direction of integrated care during the same period as our study [233]. Thus, our study addressed the question if investing in more comprehensive DMPs is cost-effective compared to less comprehensive DMPs.

Our results show that patients' physical activity was improved in all DMPs during the two-year follow-up period. This improvement was higher among patients in the most

comprehensive DMPs, except for the primary CVR-prevention sample. However, the higher improvement in the most comprehensive DMPs tends to decrease in the second year of implementation when the mean physical activity in the least comprehensive DMP increased substantially. Why this occurred remains unclear. These findings, though not statistically significant, supplement existing evidence that DMPs improve physical activity [224,234].

Moreover, the results show that in all DMPs the percentage of current smokers was reduced during the follow-up period except for CVR-DMP 9. This is in line with the findings of previous studies [224,234]. The most comprehensive DMPs appeared to be more effective in smoking cessation than the least comprehensive DMPs because they had a lower proportion of current smokers during the two-year follow-up period. This suggests that smoking cessation may be indirectly supported by interventions (e.g. monitoring, decision support systems, family participation) provided in addition to smoking specific interventions.

Furthermore, the results showed a negative trend in the quality of life of patients (expressed in QALYs) in all DMPs. The differences in QALYs between the comparators appeared to favour the most comprehensive DMPs in secondary CVR-prevention and in COPD, as 91% and 69% of bootstrap replications were in the Western quadrants, respectively. This finding could be associated with the fact that patients in these DMPs were less healthy (approximately 0.76 QALYs at baseline) than in the primary CVR-prevention and both types of CVR-prevention (approximately 0.82 QALYs at baseline). If this association exists, then comprehensive DMPs seem to be more appropriate to address the needs of more "severe" or complex patients. This is also suggested in the literature [222]. Taking into account the increase in the number of "complex" patients (e.g. patients with multi-morbidity) [235] and their substantially higher costs [236], comprehensive DMPs may be seen as means to tackle this challenge.

The yearly costs per patient from the health care perspective favoured the most comprehensive DMPs in secondary CVR-prevention and in both types of CVR-prevention. In these DMPs, patients had on average higher costs at baseline and substantially lower costs in the first and second year of implementation than patients in the least comprehensive DMPs. In contrast, patients in the most comprehensive primary CVR-prevention had higher costs from the health care perspective compared to patients in the least comprehensive DMP. However, this difference reversed when taking the societal perspective, because productivity loss was lower in the most comprehensive DMP. The costs of the COPD patients favoured the least comprehensive DMPs taking both perspectives. Taking these results into account, it can be argued that most comprehensive DMPs may be able to reduce costs, at least from the societal perspective. However, the evident variability in costs of patients included in Dutch DMPs [204,209] may influence the potential of the most comprehensive DMPs to save costs. Therefore, the comprehensiveness of

DMPs should be closely targeted to the case-mix of the patient group at interest. In addition, the potential cost savings from the implementation of comprehensive DMPs are likely to be reduced by the higher development and implementation costs associated with these programs.

An important issue of discussion is how to define the comprehensiveness of a DMP. In this study DMP comprehensiveness was defined as the number of different interventions per component of the CCM. However, previous studies showed that besides the number of interventions also the intensity of these interventions and active strategies to support implementation of these interventions were related to improved quality of care and reduced health care utilization [237,238]. Therefore, in future studies, these elements should complement the number of DMP interventions when defining the DMP comprehensiveness.

Furthermore, we found indications that the comprehensiveness of DMPs, expressed as the number of interventions, relates to higher development and implementation costs [239]. This finding should be considered thoroughly by DMP providers when designing the mix of interventions. It would be wise to estimate the comprehensiveness level at which the marginal development and implementation costs do not exceed the marginal health care savings (i.e. to find a Pareto optimal equilibrium). This estimation should anticipate that the development and implementation costs per enrolee will become lower when more patients will be enrolled in the DMP and caregivers gain experience in managing and maintaining a DMP.

The evidence provided by the CUA from the societal perspective (the perspective preferred by the Dutch health care authorities) indicated that the most comprehensive DMPs were likely to be cost saving (primary CVR-prevention, secondary CVR-prevention and both types of CVR-prevention) and/or more effective (secondary CVR-prevention and COPD) in most disease samples. Especially in secondary CVR-prevention, the most comprehensive DMP dominated the least comprehensive DMP (i.e. QALY gains at lower costs). The results from the sensitivity analysis (i.e. including development and implementation costs) were in the same line. This evidence points out that the comprehensiveness of DMPs expressed in number of interventions covering the spectrum of the CCM has the potential to improve quality of life of more severe patients (e.g. patients diagnosed with COPD and CVD) either at higher or even at lower costs.

This study has several strengths. First, the pragmatic study design allowed the evaluation of real-world data of DMPs implemented in common daily practice. Second, we selected comparators from a wide set of DMPs across four different disease categories to be compared while, most evaluation studies compare only two alternatives within a single disease. Third, different types of outcomes and costs were included in the evaluation. Fourth, we applied stabilised inverse probability weighting as a propensity score matching method to optimize comparability between DMPs. Fifth, sophisticated regres-

sion analysis was used to account for skewed data, missing responses, and repeated measurements. Sixth, the bootstrapping method applied in the analyses enabled the appropriate estimation of the uncertainty in the predicted outcomes. A major limitation of the study was the lack of clinical data. However, future research could investigate the impact of comprehensive DMPs on clinical outcomes and use decision-analytic models to extrapolate improvements in intermediate clinical outcomes to final outcomes.

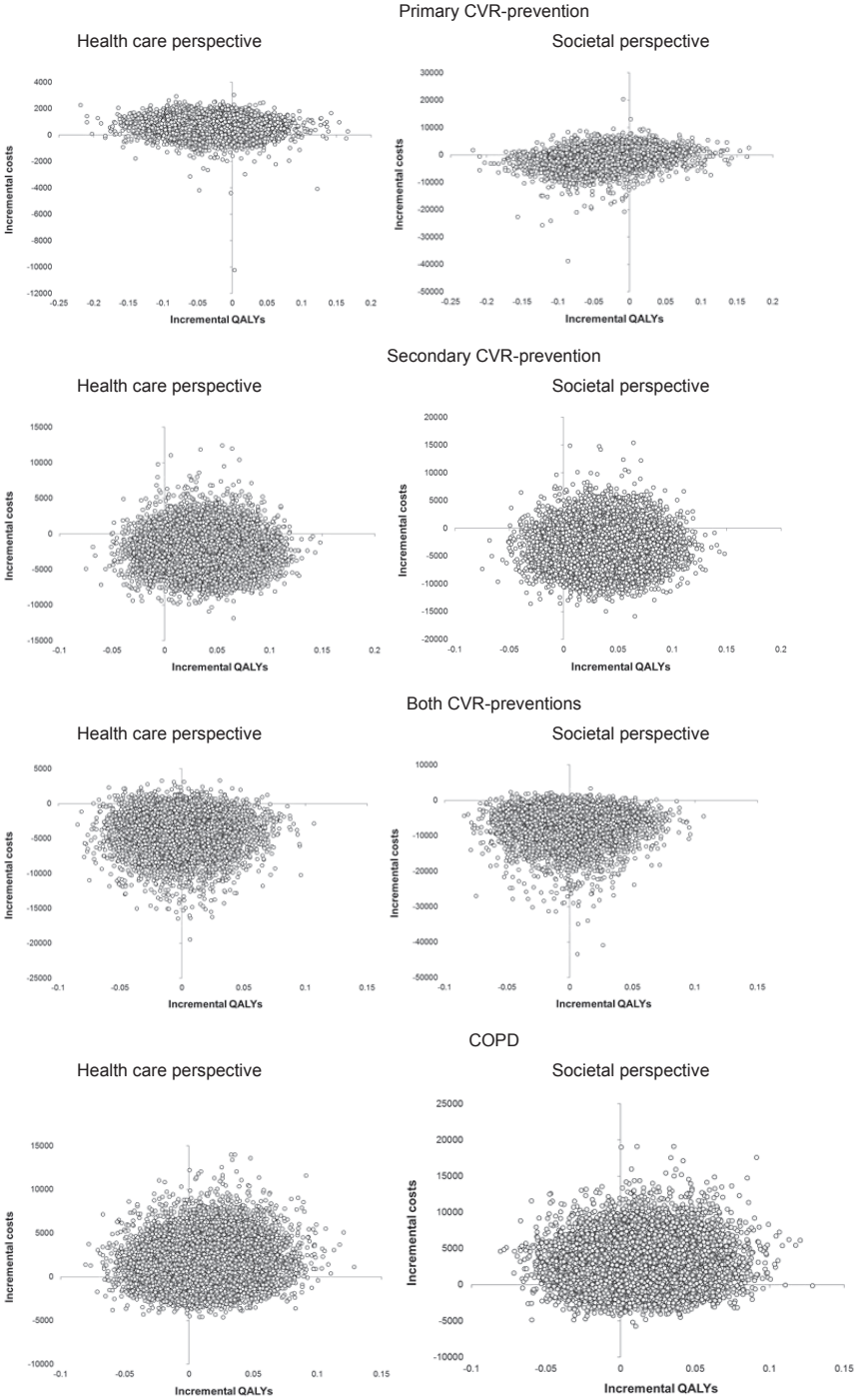
8.5 Conclusions

This study showed that developing and providing comprehensive DMPs for CVR and COPD is potentially cost-saving, effective or cost-effective. DMP providers, health insurers, and policy makers should focus on the comprehensiveness of the DMPs in the Netherlands rather than trying to identify which interventions drive the effects of integrated care. Find the right mixture of interventions that is optimally comprehensive for a specific target population is the greatest challenge.

Appendix 1 Unit cost prices used in the costs analysis

	2009	2010	2011	2012
Inflation rate- Dutch Statistics (CBS)	1,2	1,3	2,3	2,5
GP session	28	28	29	30
GP home visit	43	44	45	46
GP phone contact	14	14	15	15
Inpatient day	457	463	474	485
Day care treatment	251	254	260	267
Intensive care unit day	2,183	2,211	2,262	2,319
Outpatient visit	72	73	75	76
Emergency room visit	151	153	156	160
Physical therapy	36	36	37	38
Speech therapy (session)	33	33	34	35
Occupational therapy (hour)	22	22	23	23
Dietary advice (hour)	27	27	28	29
Home care (hour)	35	35	36	37
cost/km	0.20	0.20	0.21	0.21

Appendix 2 Cost-effectiveness planes from the main adjusted CUA



Appendix 3 Observed mean costs and mean QALYs per DMP

			Total health care costs		**Total societal costs**		**QALYs**	
			1st Year	2nd Year	1st Year	2nd Year	1st Year	2nd Year
Primary CVR-prevention	CVR-DMP 4	mean	1,197	1,250	1,888	1,588	0.825	0.811
		SD	(1,703)	(2,256)	(6,834)	(6,186)	(0.172)	(0.151)
		n	83	63	121	121	77	41
	CVR-DMP 7	mean	969	1,001	3,813	841	0.829	0.728
		SD	(1,197)	(1,358)	(11,966)	(1,381)	(0.172)	(0.292)
		n	20	20	30	30	18	9
Secondary CVR-prevention	CVR-DMP 3	mean	2,574	2,727	3,916	2,891	0.791	0.810
		SD	(5,242)	(8,720)	(10,456)	(8,735)	(0.181)	(0.097)
		n	58	33	58	33	56	26
	CVR-DMP 1	mean	4,795	3,752	5,164	5,224	0.749	0.704
		SD	(11,485)	(10,629)	(12,044)	(12,499)	(0.197)	(0.179)
		n	78	57	78	57	70	39
Both types of CVR-prevention	CVR-DMP 6	mean	2,897	1,638	3947	1929	0.810	0.762
		SD	(15,988)	(2,724)	(16998)	(3292)	(0.190)	(0.213)
		n	159	145	159	145	151	96
	CVR-DMP 9	mean	4,270	4,900	6,220	6,275	0.799	0.755
		SD	(7,724)	(21,398)	(14,906)	(22,807)	(0.208)	(0.220)
		n	90	102	90	102	79	53
COPD	COPD-DMP 4	mean	4,556	2,491	5,399	7,016	0.806	0.749
		SD	(9,309)	(2,404)	(10,596)	(14,357)	(0.180)	(0.233)
		n	65	67	65	78	63	49
	COPD-DMP 1	mean	6,010	3,663	2,654	3,957	0.689	0.671
		SD	(12,695)	(4,888)	(2,554)	(5,151)	(0.219)	(0.227)
		n	78	81	67	81	77	56

CHAPTER 9

Broader economic evaluation of disease management programs using multi-criteria decision analysis

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Abstract

Objectives: The aim of this paper is to develop a methodological framework to facilitate the application of Multi-Criteria Decision Analysis (MCDA) for a comprehensive economic evaluation of disease management programs (DMPs).
Methods: We studied previously developed frameworks for the evaluation of DMPs and different methods of MCDA and we used practical field experience in the economic evaluation of DMPs and personal discussions with stakeholders in chronic care.
Results: The framework includes different objectives and criteria that are relevant for the evaluation of DMPs, indicators that can be used to measure how DMPs perform on these criteria, and distinguishes between the development and implementation phase of DMPs. The objectives of DMPs are categorised into a) changes in the process of care delivery, b) changes in patient lifestyle and self-management behaviour, c) changes in biomedical, physiological and clinical health outcomes, d) changes in health-related quality of life, and e) changes in final health outcomes. All relevant costs of DMPs are also included in the framework. Based on this framework we conducted a MCDA of a hypothetical DMP versus usual care.
Conclusions: We call for a comprehensive economic evaluation of DMPs that is not just based on a single criterion but takes into account multiple relevant criteria simultaneously. The framework we presented here is a step towards standardising such an evaluation.

9.1 Introduction

Many countries have taken initiatives toward integration in chronic care by developing and implementing disease management programs (DMPs), which are expected to improve effectiveness and efficiency of chronic care delivery [47,57]. Schrijvers (2009) define these programs as "a group of coherent interventions designed to prevent or manage one or more chronic conditions using a systematic, multidisciplinary approach and potentially using multiple treatment modalities" [24]. Health technology assessment (HTA) could have a leading role in informing decision makers around the world about the extent to which DMPs meet these expectations [49]. However, despite the attention that DMPs have received the last decade, the evidence from the relatively few HTA studies [2,49] is inconclusive. This can largely be explained by the variation in design, outcome measures, and costing methods used in the economic evaluations of DMPs [57,62].

Evidentially, the lack of a methodological framework for the HTA of DMPs has contributed to this variation and made the comparison of results hardly possible, meaning that much time and financial resources have been spent inefficiently [63]. Current decision-making incorporates traditional cost-effectiveness studies that may not be suitable for the comparison between DMPs and usual care. This is because DMPs are complex, multifaceted interventions that have multiple effects such as improved self-management capabilities, coordination and continuity of care, reduced risk factors and complication rates, improved quality of life, which cannot be expressed in a single unit of effect like a quality-adjusted life-year (QALY) that is traditionally used in economic evaluations of health care interventions. Therefore, the establishment of a methodological framework to perform an analysis incorporating the most relevant costs and effects is desirable in performing economic evaluations of DMPs valuable to decision making.

Multi-Criteria Decision Analysis (MCDA) is developed to support decision making by allowing for a systematic trade-off between multiple, and sometimes conflicting effects and costs simultaneously in an explicit, transparent and consistent way [240,241]. Usually, policy makers implicitly consider and weigh those outcomes (criteria) and incorporate them in decision-making in a deliberate way. However, such "intuitive" decision making may not be transparent and it may be complicated by conflicting criteria or different opinions among stakeholders regarding the importance of different criteria [242], which jeopardizes the accountability of decision makers to patients, insurance-payers, and professionals.

The aim of this study is to develop a methodological framework to facilitate the application of MCDA in a broader economic evaluation of DMPs including the most relevant outcomes and cost categories.

9.2 Methods

We developed a methodological framework for the application of MCDA in a large study in which we evaluate twenty-two DMPs for several chronic diseases in the Netherlands [128]. For the development of the framework, we studied literature to identify and understand MCDA techniques that could be applied to the evaluation of DMPs. To identify the objectives and criteria—the first and most important steps in MCDA—we studied frameworks that had previously been used to evaluate DMPs. The frameworks of Steuten et al. [63] and Lemmens et al. [243] were the most relevant in this respect. We also used practical field experience in the ongoing broad HTA of the twenty-two DMPs mentioned above and personal discussions with integrated care providers, health insurers, scientific and practical experts in the integrated chronic care field as inspiration in understanding the complexities of DMPs and conceiving the idea for the application of MCDA in their evaluation. Finally, we conducted a MCDA of a hypothetical DMP versus usual care based on our framework to illustrate its application.

9.3 An introduction to MCDA

MCDA has been successfully applied in other areas of public decision making, like the environmental area [244]. Interest in MCDA for priority-setting in health care is growing rapidly the last decade [245-247]. This is because MCDA overcomes the limitations of other priority-setting techniques such as cost-effectiveness, burden of disease, or equity analysis that concentrate on single criteria [248]. MCDA elicits preferences for alternative interventions by assessing the extent to which the objectives have been achieved using measurable criteria [245]. In this process, different criteria are weighted according to their relative importance to the decision. Hence, MCDA is a sophisticated method for comparing complex interventions such as DMPs incorporating all relevant categories of outcomes and costs [247,249,250].

The mains steps in conducting a MCDA include: (i) establishing the decision context and identifying the options to be appraised, (ii) identifying objectives and criteria, (iii) scoring by measuring the performance of each option on each criterion, (iv) assigning weights to each criterion, (v) combining the weights and scores to get the overall value, (vi) examining the results and performing sensitivity analysis [246]. There are numerous different techniques for performing MCDA and their selection depends on the decision situation and the familiarity of the researchers/decision makers with a MCDA technique. However, the techniques that have proven to be most feasible and suitable are the Multi-Attribute Value Theory (MAVT) and the Analytic Hierarchy Process (AHP) [244].

9.4 Evaluation frameworks relevant to DMPs

Steuten et al. [63] and Lemmens et al. [243] developed frameworks to evaluate DMPs. These frameworks identified structure, process, and outcome indicators used in the evaluation of DMPs. The structure indicators (e.g., method of reimbursement, presence of ICT system) are not relevant to HTA because they cannot be used to assess and quantify the performance of a DMP. Rather, these indicators are conditions for a DMP to perform well influencing therefore the process and outcome indicators. Lemmens et al. [243] distinguished two mechanisms underlying the effects of DMPs on processes and final outcomes. The first is the patient's learning and behavioral change mechanism and the second is the professional support and behavioral change mechanism. These mechanisms lead to changes in process indicators such as disease-specific knowledge and self-care behavior as well as adherence to evidence-based guidelines and use of monitoring systems, respectively. Both frameworks relate changes in processes to changes in outcomes such as health-related quality of life (HR-QoL), mortality, clinical health status, and all relevant costs and distinguish them as important factors in the evaluation of DMPs.

9.5 Identifying objectives, criteria, and measurements

Using the previously mentioned frameworks, we identified objectives of DMPs that can be included in the second step of performing a MCDA. The extent to which these objectives are achieved can be assessed by introducing a set of criteria similar to the process and outcomes indicators included in the frameworks of Steuten et al. [63] and Lemmens et al. [243]. In the next sections, we discuss different criteria per objective and we provide some examples of indicators that could be used to measure the performance scores on each criterion (step 3 in MCDA).

9.5.1 Criteria to Assess the Performance of DMPs

The effects of DMPs cover a wider range of outcomes influencing aspects of the delivery process as well as intermediate and final health outcomes. Although the ultimate objective might be to improve health outcomes, it should be kept in mind that it may take a long time before quality improvements in structure and process are translated into changes in health outcomes [53]. Thus changes in the process of care delivery and changes in intermediate outcomes may become goals by themselves.

9.5.2 Changes in the Process of Care Delivery

Because one of the main objectives of DMPs is to change care delivery toward integration of care, measurements of this process change should be used. The Assessment of Chronic Illness Care (ACIC) and the Patient Assessment of Chronic Illness Care (PACIC) could be used as a process indicator of integrated care improvement from the care professional and patient perspective, respectively [25]. These instruments cover many process indicators described in the framework related to performance of care providers and continuity of care by Steuten et al. [63]. Moreover, as co-ordination between professionals of different disciplines is a crucial element of effective disease management [251], measurements of coordination level such as the relational coordination survey [252] could be used in the evaluation. In addition, performance indicators, such as proportion of patients receiving care according to evidence-based guidelines would also be suitable to measure changes in care delivery [253]. Examples are the proportion of participants that get smoking cessation support in a COPD-DMP, the proportion of participants receiving podiatric care and annual eye controls in a diabetes-DMP, and the proportion of participants receiving statins in a cardiovascular-DMP.

9.5.3 Changes in Patient Lifestyle and Self-management Behavior

Because lifestyle improvement of people with chronic conditions is an important objective of DMPs, measurements of patients' lifestyle behavior such as smoking, exercise and nutrition should be part of the evaluation [68]. There are numerous instruments to measure physical activity, including self-report questionnaires such as the Epic Norfolk Physical Activity Questionnaire (EPAQ) [254] and activity monitors like pedometers and accelerometers. The lifestyle changes are part of self-management. But self-management includes much more than this. It refers to any behavioral change that enables patients to take conscious decisions on many aspects of every-day life with a chronic disease. It includes accepting the disease, maintaining social contacts and support, keeping emotional balance, exercises to improve self-efficacy and adaption to the disease, for example by applying energy-saving techniques, stress management, working on adequate illness perceptions, etc. It also refers to teaching patients to adequately comply with therapy and how to act in case of disease worsening [68]. Part of this is measured by the Self-Management Ability Scale (SMAS) [155].

9.5.4 Changes in Biomedical, Physiological, and Clinical Health Outcomes

Depending on the disease that is targeted, changes in biomedical, physiological, and clinical health outcomes such as blood pressure, cholesterol, forced expiratory volume in 1 second (FEV1), glycated hemoglobin (Hba1c), exacerbations, and complications are crucial outcome measurements of DMPs, because they may change disease progression and predict long-term changes in the health status of a patient. These outcomes are

also informative to providers and contractors of DMPs (e.g., health insurers) that are negotiating about the quantity, price, and quality of care [47,196].

9.5.5 Changes in Health-Related Quality of Life

Health-related quality of life should be incorporated in the evaluation of DMPs to assess improvements in the quality of life of the participating patients. Although disease-specific questionnaires may be more sensitive to change, it can be argued that generic measurements of HR-QoL such as the Short Form 36 (SF-36) or the EuroQol 5 dimensions questionnaire (EQ-5D) might be most suitable to the evaluation of DMPs because a significant proportion of patients with a chronic disease suffers from multiple morbidities. A DMP targeted to one disease may have spill-over effects on other diseases (e.g., multiple diseases may benefit from more physical activity or a better nutritional status). However, disease-specific measures such as the St George's Respiratory Questionnaire (SGRQ) and domain-specific measures such as the Barthel scale (measuring activities of daily-living) [255] could also be included, depending on the purposes of the study.

9.5.6 Changes in Final Health Outcomes

The time horizon of empirical (economic) evaluations of DMPs is often not long enough to actually observe a change in (quality-adjusted) life-years. When the association between previously mentioned categories of health outcomes and the changes in quality and length of life is clear, it may be possible to extrapolate the outcomes that occur within a shorter time period into life-years or QALYs gained using decision analytic disease models [53]. Although there are some attempts to include self-management and patient perceptions in such models [256], extensive applications suitable for DMPs do not exist yet.

9.5.7 Related Costs

The costs in the evaluation of the DMPs can be distinguished into direct costs within the health care sector: (i) costs of development, (ii) costs of implementation, (iii) costs of diagnosis and treatment, direct costs outside the health care sector: (iv) costs borne by the patient/family, (v) costs of informal care, and indirect costs: (vi) costs of productivity losses (vii). Measurements of health care usage costs (e.g., outpatient and inpatient care and medication costs), the costs borne by the patient/family, the costs of informal care, and costs of productivity loss are similar to the conventional medical technologies, and are extensively discussed in the literature [255]. Therefore, we discuss only the measurement of development and implementation costs hereafter.

9.5.8 Development and Implementation Costs

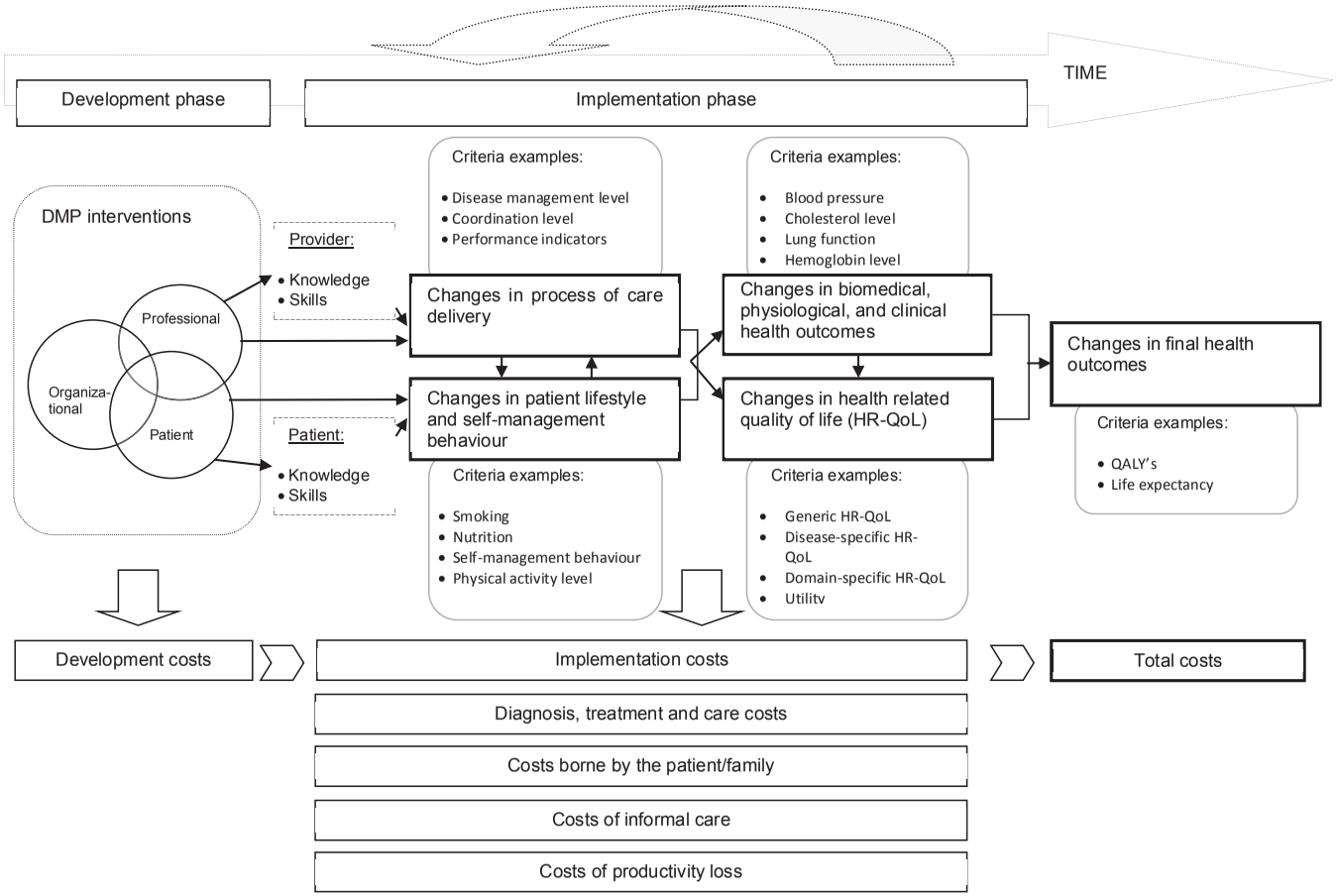
Development costs include all costs made during the preparation phase of a DMP such as labor time of personnel that participated in brainstorming sessions and logistic arrangements, training costs, and costs of software that supports audit and feedback. These costs could be estimated by using information that can generally be obtained from the managers of DMPs. The DMP implementation costs begin when the provision of DMP interventions to patients starts. Examples of implementation costs include the costs of managing the DMP, the costs of multidisciplinary team meetings, the costs associated with collecting quality of care indicators for audit and feedback, the costs of materials used for patient education, and the costs of keeping the ICT operating. Costing instruments such as the one developed by the World Health Organization [197] could be adjusted to systematically collect the development and implementation costs of DMPs.

9.6 Assembling the framework for the MCDA of DMPs

Having identified the most important objectives and criteria for assessing the achievement of each objective, we developed a framework for the application of MCDA in the economic evaluation of DMPs. The framework (see Figure 1) distinguishes between the development phase of DMPs and the implementation phase. In the development phase, a mixture of patient-directed (e.g., self-management training), professional-directed (e.g., education and training) and organizational interventions (e.g., electronic patient records) [243], are usually selected, designed, and prepared to be implemented. The development costs accumulated in this phase are also incorporated in our framework.

In the implementation phase, the interventions are infused in the organization that provides the DMP to patients with a single or multiple chronic diseases. One of the short-term outcomes can be a change in patients' and providers' knowledge, skills, and attitudes that can have an impact on the uptake of patient/professional-directed interventions and influence changes in other outcome measures [243]. Hence, in our framework, the changes in knowledge, skills and attitudes are seen as part of the mechanism through which the changes in the outcome categories can be achieved, rather than as separate outcome categories themselves. The framework shows that DMP interventions can aim to influence the process of care delivery and patient lifestyle and self-management behavior directly or by means of professional and patient knowledge, skills, and attitude. Suggested criteria to assess the changes in the patient lifestyle and self-management behavior are also part of the framework. They include self-management abilities, smoking behavior, physical activity, and nutrition. Likewise, the framework includes suggestions for criteria to assess changes in the care delivery process, for example disease management level, coordination level and indicators of the

Figure 1 Framework for the economic evaluation of DMPs



extent to which care is provided according to guidelines/care standards. In many cases, changes in the process of care delivery would trigger changes in patient behavior and vice versa.

Both changes in patient behavior and care delivery lead to changes in biomedical, physiological, and clinical health outcomes and changes in HR-QoL. Furthermore, changes in biomedical, physiological, and clinical health outcomes may influence the HR-QoL. Together, these changes result in changes in final health outcomes (QALYs and/or life expectancy) in the medium-term, but more likely, the long-term.

We acknowledge that disease management is an iterative process in which changes in biomedical, physiological and clinical outcomes and change in HR-QoL may trigger new changes in the process of care delivery and patient behavior. This is indicated in our framework by the circular arrow at the top of Figure 1.

Finally, the costs that occur during the implementation phase (i.e., DMP implementation costs, treatment costs, costs borne by the patient/family, costs of informal care, and costs of productivity loss) are added to the framework to ensure the calculation of the total costs of the DMPs.

9.7 A hypothetical case study

To illustrate how MCDA can be performed in the evaluation of DMPs, a hypothetical example is given of a COPD-DMP that is compared with usual care. Usual care is defined as "care most commonly provided by organizations without a DMP" [62]. A list of interventions such as included in Supplementary Table 1, may be used as a checklist to distinguish between the interventions in usual care and the interventions in the DMP. The DMP aims to achieve improvements in all outcome categories of the framework (i.e., the six bold squares) plus a measure that relates the costs to the final outcomes, that is, the cost-effectiveness. For each objective, one or more criteria are chosen from the framework to compare the performance of the two treatments.

In this example, for changes in process of care delivery the self-management support criterion (measured by the ACIC) is chosen. Smoking and self-efficacy score are chosen as criteria for changes in patient lifestyle and self-management. They were measured as the percentage of patients that had successfully quit smoking at 12 months and by the self-efficacy domain of the SMAS, respectively. The criterion for the biomedical health outcomes is the lung function measured in FEV1% predicted and for changes in HR-QoL is the disease-specific QoL measured by the SGRQ. Moreover, the total costs per patient are selected as the criterion for the costs and the QALY is the criterion for changes in final health outcomes in this example. In addition to these criteria, the cost-effectiveness ratio calculated by dividing total costs per patient by the QALY per patient is also taken

Table 1 Example of a performance matrix

Objective	Changes in process of care delivery	Changes in patient lifestyle and self-management		Changes in biomedical, physiological, and clinical health outcomes	Changes in health related quality of life	Total costs	Changes in final health outcomes	Combination of objectives
Criterion	Self-management support	Smoking	Self-efficacy	Lung function	Disease specific quality of life	Total costs per patient	QALY	Cost-effectiveness ratio
measurement	ACIC domain self-management	% quit smoking after 12 months	SMAS domain self-efficacy	FEV1% predicted	SGRQ	-		Costs/ QALY
range	0-11 (best)	0-100 (best)	1-6 (best)	0-100 (best)	0-100 (worst)	0-∞ (worst)	0-∞ (worst)	0-∞ (worst)
DMP	7.65**	22%*	4.23*	75.4%**	37#	€13,565#	1.62#	€8,373
usual care	3.81**	23%*	4.24*	76%**	40#	€10,814#	1.54#	€7,022

*Source: [245], ** Source: [247]; # Source: [128]; Note: Total costs per patient and QALYs are estimated in two-years period

into account. To bring our hypothetical DMP closer to reality, we have used findings from existing studies that evaluated DMPs and usual care against these criteria. We used three studies [127,197,257] as we could not find a single study reporting results for the whole spectrum of the selected criteria. The results are shown in the performance matrix in Table 1.

The next step in implementing MCDA is to standardize the performance measures (i.e., retransform the results on different criteria onto the same scale). In this example, we have chosen a method that enables us to standardize performance measures with different ranges which is:

$$S_{ij} = \frac{x_{ij}}{(x_{ij}^2 + x_{yj}^2)^{1/2}}$$

where S_{ij} is the standardization of the performance value x of the i alternative against the j criterion, where i is either the DMP or usual care and j are the criteria in Table 1. The performance value of the y alternative against the j criterion is denoted as x_{yj}. In the case of the SQRG scores and costs where the scale has a reverse direction (the highest the least preferred), the reciprocal of the performance values (i.e. $1/x_{ij}$ and $1/x_{yj}$) is used in the formula. After standardization, the performance values have a range between 0 (lowest) and 1 (highest). The standardized performance values of each criterion for the two alternative options are presented in Table 2.

Following the steps of the MCDA, weights have been attached to each criterion in our example. These weights reflect the relative importance of each criterion in the decision making. As mentioned in a previous section, there are several methods to obtain these weights but their application is outside the scope of this paper. Complying with MCDA manuals, the sum of the hypothetical weights is 1 [249].

In the next step the standardized performance values are combined with the criteria weights in order to estimate the total scores. These are calculated using:

$$T_i = \sum_{j=1}^{n} S_{ij} \times w_j$$

where T_i is the total score for alternative i and w_j is the weight for criterion j. As illustrated in Table 2, the total score is 0.73 for the hypothetical DMP and 0.67 for usual care. Considering this, the DMP option is preferred to the usual care option because it has the highest total score in our example.

Table 2 Example of scoring the two treatment alternatives

Criterion	Self-management support	Smoking	Self-efficacy	Lung function	Disease specific quality of life	Total costs per patient	QALY	Cost-effectiveness ratio	Total score*
Weights	0.20	0.10	0.08	0.09	0.10	0.08	0.15	0.20	1.00
Standardized performance scores									
DMP	0.90	0.69	0.71	0.70	0.73	0.62	0.72	0.64	0.73
Usual care	0.45	0.72	0.71	0.71	0.68	0.78	0.69	0.77	0.67

* Total score was calculated as a weighted sum of the standardized performance scores

9.8 Discussion and conclusion

Our framework contributes to the methodology of HTA of DMPs by providing an analytical structure to set up a MCDA in the complex field of disease management where multiple comparisons of different interventions and different outcomes and costs have to be made simultaneously. By valuing a broader set of outcomes than just the QALY, this method may overcome the limitations of a conventional CEA. Moreover, even when a DMP evaluation has a time horizon that is not long enough to capture changes in final health outcomes, this framework can still support decision making because it considers a great range of objectives and outcomes. This framework can also evaluate DMPs targeted to patients with multi-morbidity by selecting criteria that are relevant for such population.

It is important to note that our framework does not intend to explain the mechanisms through which changes can be achieved, as it is done by many theories on behavioral change. Our framework focuses on the decision criteria used in evaluating a DMP. Nevertheless, one of the major challenges is the selection of the criteria to be included in the MCDA, because it is impossible to include all aspects that are possibly influencing decision making. Should they be restricted to the objectives (outcomes categories and costs) included in our framework or are there other, wider criteria that need to be incorporated such as size of the target population or the difficulty to motivate the target population? Should the average cost/QALY ratio be one of the criteria or not, considering that decision making is commonly based on the incremental ratio of additional costs compared with current care divided by the gain in QALYs? Also, for the same objective, different and multiple criteria can be chosen, which may be equally relevant and for each criterion multiple indicators and measurements may be available. To overcome these challenges, previous studies have mostly used literature reviews, semi-structured interviews, and expert opinions to restrict the criteria to a manageable number [247,258]. One way forward would be that the researchers and decision makers would agree on a core or minimal set of criteria and indicators for each objective that

would be used for a certain period (see Supplementary Table 2). Additional criteria could be added to this set where relevant.

Obtaining the weights is another challenge. There are different methods available, which are roughly categorized into value-based methods, outranking methods, and goal-achievement methods [244]. A discrete-choice experiment is an example of a value-based method, but it requires independence between the criteria. The application of the Analytic Network Process (ANP), which is an extension of the AHP, may be the most suitable as it overcomes the concerns about the dependence between criteria [259]. AHP is a value-based approach using pair-wise comparisons between criteria and DMPs to derive numerical weights and performance scores. It is called hierarchical because criteria can be divided into sub-criteria. The scores and weights are developed for each individual criterion initially and then aggregated assuming multiplicative preferences. The essence of ANP is the possibility to include dependence between the criteria in a decision. This advantage seems to be important to perform an evaluation of DMPs using MCDA because the outcomes of DMPs may be interacting, failing therefore, to ensure independence between the criteria, as it is required by the other MCDA methods.

In conclusion, we have presented a framework for the application of MCDA to simultaneously assess the broader outcomes and costs of DMPs. This methodology may stimulate and facilitate a much broader economic evaluation of DMPs that is currently done. It is desirable to further explore the applicability of MCDA approaches to DMPs. Therefore, we have planned empirical applications of this framework within the context of a large study in which we evaluate twenty-two different DMPs [128]. The framework could be used in reimbursement decisions for DMPs or in negotiation processes between DMP providers and health insurers after having collected the necessary information on the selected criteria. Using this framework, decision makers on governmental and organizational level as well as health insurers and other payers could be provided with comprehensive information about what DMPs actually deliver on patient, professional caregiver, and organizational level and to what costs. This would improve the transparency about which criteria play a role in the decision making process and to what extent [242]. As a result, the results of MCDA could support decision makers to improve consistency in decision making and accountability to patients and professionals with the final aim to improve the quality and efficiency of chronic disease care.

Supplementary Table 1 Indicative list of DMP interventions per Chronic Care Model component*

Interventions per CCM component	Usual care (√)	DMP (√)	Description of specific intervention
Organizational support			
Integrated financing			
Specific subsidies for foreign population			
Sustainable financing agreements with health insurers			
Other:			
Community			
Cooperation with external community partners			
Treatment and care pathways in outpatient and inpatient care			
Involvement of patient groups and patient panels in care design			
Discussion panel for community partners related to chronic care			
Regional training course			
Family participation			
Other:			
Self-management			
Promotion of disease specific information			
Individual care plan			
Life-style interventions (e.g. physical activity, diet, smoking cessation)			
Support of self-management (e.g. internet, email or sms, e-consultation)			
Tele-monitoring			
Personal coaching			
Motivational interviewing			
Informational meetings			
Mirror interviews			
Group sessions for patient and family			
Cognitive behavioural therapy			
Other:			
Decision support			
Care standards			
Uniform treatment protocol in outpatient and inpatient care			
Training and independence of practise assistants			
Professional education and training for care providers			
Audit and feedback			
Development and implementation of care protocols for immigrants			
Structural participation in training sessions			
Quality of Life questionnaire			
Registration of process and outcome indicators			
Qualitative evaluation of health care via focus-groups with patients			

Interventions per CCM component	Usual care (√)	DMP (√)	Description of specific intervention
Periodic evaluation of DMP interventions and feedback			
Measurement of patient satisfaction			
Other:			
Delivery system design			
Multidisciplinary cooperation between outpatient and inpatient care			
Delegation of care from specialist to nurse/care practitioner			
Development of health pathways and protocols			
Substitution of inpatient with outpatient care			
Systematic follow-up of patients			
Specific plan for immigrant population			
Meetings of different disciplines for exchanging knowledge/ information			
Monitoring of high-risk patients			
Board of clients			
Periodic discussion sessions between care professionals and patients			
Stepped care method			
Other:			
ICT			
Electronic Patient Records system (with/without patient portal)			
Hospital Information System			
Integrated Information System			
Use of ICT for Internal and/or regional benchmarking			
Systematic registration by every caregiver			
Exchange of information between different care disciplines			
Other:			

adjusted from: Wagner, E. H., Glasgow, R. E., Davis, C., Bonomi, A. E., Provost, L., McCulloch, D., et al. (2001). Quality improvement in chronic illness care: A collaborative approach. *The Joint Commission Journal on Quality Improvement, 27*(2), 63-80. *Wagner's Chronic Care Model (CCM) provides a framework of elements that must be considered when developing improvement strategies for providing care for people with chronic diseases, originally including: a) self-management, b) decision support, c) delivery system design, d) clinical information systems, e) health care organization, and f) community resources and policies.

Supplementary Table 2 Indicative list of core criteria and potential indicators per objective

Changes in process of care delivery	
Criteria:	Indicators:
Disease management level	Assessment of Chronic Illness Care (ACIC), Patient Assessment of Chronic Illness Care (PACIC)
Coordination level	Relation coordination survey
Performance indicators	Indicators related to care process e.g. proportion of patients receiving care according to evidence-based guidelines
Changes in patient lifestyle and self-management behaviour	
Criteria:	Indicators:
Smoking	% of participant that quitted smoking
Nutrition	Nutritional status e.g. % of daily calories available from fat, Body Mass index
Self –management behaviour	Self-Management Ability Scale (SMAS)
Physical activity level	Epic Norfolk Physical Activity Questionnaire (EPAQ), Short Questionnaire to Assess Health-enhancing Physical Activity (SQUASH), activity monitors (e.g. pedometers and accelerometers)
Changes in biomedical, physiological, and clinical health outcomes	
Criteria (depending on disease(s) of the target population):	Indicators:
Blood pressure	mmHg
Cholesterol level	mmol/l
Lung function	FEV_1, FVC
Changes in health related quality of life (HRQoL)	
Criteria:	Indicators:
Generic HRQoL	SF-36 questionnaire
Utilities	EQ-5D questionnaire, SF-6D questionnaire
Changes in final health outcomes	
Criteria:	Indicators:
Quality Adjusted Life Years	QALYs
Changes in costs	
Criteria:	Indicators:
Costs from the health care perspective	Sum of: a) developments costs, b) implementation costs, c) diagnosis, treatment and care costs
Costs from the societal perspective	Sum of the above mentioned costs plus: a) costs borne by the patient/family, b) costs of informal care, c) costs of productivity loss
Changes in combined objectives	
Cost-effectiveness/utility	Incremental cost-effectiveness/utility ratio

CHAPTER 10

General discussion

10.1 Main findings

10.1.1 Payment schemes implemented to promote integrated care and their impact

Several European countries have implemented financial agreements to promote the integration of chronic care, including pay-for-coordination (PFC), pay-for-performance (PFP), bundled payment and global payment. These agreements provided incentives to different stakeholders. The reforms in Austria (financial pooling), Denmark (regional budgets), and Germany (global payment) provided financial incentives and means to financial poolers and payers. On the other hand, the implementation of PFP in France, England, Estonia, Hungary and Portugal targeted the financial reward of primary care physicians. In the Netherlands however, the implementation of bundled payment provided financial incentives to both health insurers and health care providers. Combinations of financial agreements were evident in France (PFC and PFP) and Germany (Global payment and PFC).

The interviews in Chapter 2 showed that all financial agreements appeared to have changed the structure of chronic care delivery. PFC, as it was implemented in Austria, France and Germany, was perceived to be the most successful in increasing collaboration within and across health care sectors, whereas PFP, as it was implemented in England and France, was perceived most successful in improving other indicators of the quality of the care process. The most commonly mentioned barriers to the implementation of payment schemes were the misaligned incentives among stakeholders and gaming of the system by care providers.

In the Netherlands, these barriers could be overcome by meeting a set of necessary conditions described in Chapter 3. These conditions included written liabilities and responsibilities of stakeholders that are linked to care standards, promotion of financial incentives to health insurers to act as purchasers of efficient chronic care, establishment of transparent relationship between performance indicators and the bundled payment and development of adequate information systems.

Regarding the impact of the payment schemes, the results from the empirical analysis in Chapter 4 showed that PFP and all-inclusive financial agreements have the potential to reduce substantially the growth of health care expenditure. It also appeared that PFC has the potential to reduce outpatient health care expenditure. In details, the annual growth of outpatient expenditure was decreased in countries with PFC (by 21.28 US$ per capita) and in countries with all-inclusive agreements (by 216.60 US$ per capita). When modeling impact as a non-linear function of time during the total 4-year period after implementation, PFP decreased the growth of hospital and administrative expenditure and all-inclusive agreements reduced the growth of outpatient expenditure.

10.1.2 Variability in costs related to DMPs

The results in Chapter 5 demonstrated the existence of large variability in costs of patients with different chronic diseases enrolled in Dutch DMPs. This variation ranged across the investigated DMPs from about €400 to €2,000 (including travelling and productivity loss) per patient per 3-months and was present within and across diseases. The results further showed that age, the presence of cardiovascular disease, multi-morbidity and payments on top of the payment for the usual care had a positive relation with costs, while better quality of life was associated with lower health care costs.

Further, the results in Chapter 6 demonstrated large variability in the development and implementation (D&I) costs of DMPs implemented in the Netherlands. The development costs varied from €5,891 to €274,783 and the implementation costs varied from €7,278 to €387,879 across DMPs. This variation can be explained by the large variability in DMP development duration, size of DMP providing organization, and the level of integrated care in the providing organization prior to the implementation of a DMP. The qualitative analysis showed that the development duration and level of integrated care were associated with the attributes of the interventions, project leadership, and the history of the ICT systems used in a DMP. The findings also indicated the existence of economies of scale and economies of scope which may reduce D&I costs.

10.1.3 Cost and effects of DMPs

After a year of implementation (Chapter 7), DMPs were associated with improvements in integration of CVR care (0.10 PACIC units), physical activity (+0.34 week-days in which patients had at least 30 minutes of moderate or severe physical activity) and smoking cessation (8% less smokers) in all diseases. Furthermore, increases in physical activity and in self-efficacy were predictive of an improvement in patient's quality-of-life. On the cost side, the changes in health care utilization costs were not statistically significant and the D&I costs were an important driver of total costs per patient included in a DMP. When comparing the most with the least effective DMP in a disease category, the vast majority of bootstrap replications (range:73%;97%) pointed to cost savings, except for COPD (21%). QALY gains were small (range:0.003;+0.013) and surrounded by great uncertainty.

Moreover, the results from the two-year follow-up after DMP implementation (Chapter 8) also showed that patients' physical activity was improved and the percentage of current smokers was reduced in (almost) all DMPs. However, patients in the most comprehensive DMPs increased their physical activity more (except for the primary CVR-prevention) and had higher smoking cessation rates. In addition, the evidence provided by the CUA from the societal perspective (the perspective preferred by the Dutch health care authorities) indicated that the most comprehensive DMPs were likely to be cost saving (primary CVR-prevention, secondary CVR-prevention and both types of

CVR-prevention) and/or more effective (secondary CVR-prevention and COPD) in most disease samples.

Although Chapters 7 and 8 included a broad range of costs and effects in the evaluation of DMPs, the application of the framework provided in Chapter 9 would have further strengthen the broad and robust evaluation of DMPs. The developed framework facilitates the application of multi-criteria decision analysis (MCDA) by including different objectives and criteria that are relevant in the evaluation of DMPs, providing indicators to measure the performance of DMPs, and distinguishing between the development and implementation phase of DMPs.

10.2 Designing appropriate payment schemes for integrated care in Europe

During the last three decades, purchasers of health care in the U.S. have investigated alternative payments schemes to stimulate integration of care. These schemes aimed to contain costs and improve quality of care. The most well-known payment schemes implemented in the U.S. are global payment, accountable care organizations with shared savings program, PFP, bundled payment, and PFC [71]. These schemes are not just an alternative method for paying health care providers. They introduced financial risk to providers, explicit measures of quality improvement driven by financial incentives to providers, efforts towards patient-centred care through integration and coordination of care, and financial incentives for patient safety. Positive evidence from the implementation of these payment schemes are reported in the literature [44,46,166,260].

Part A of this thesis showed that most the abovementioned payment schemes were adopted in European countries to reach the same aims (i.e. cost containment and improved quality of care) by stimulating integrated care. However, most of these payment schemes were adapted to be "transferable" to the European context and were accompanied by broader reforms in the European health care systems. The adoption (i.e. which) and adaptation (i.e. how) of a payment scheme in a European country were based on the structure of its health care system. For example, all-inclusive payments (i.e. bundled payment and global payment) were adopted in countries with a risk-equalization system in health care financing (e.g. the Netherlands and Germany). Further, payment schemes were often combined (e.g. global payment with PFC in Germany) or provided on top of traditional payment schemes (e.g. PFP on top of capitation and FFS in England). Using the main features of a health care system as stepping stones to achieve a payment reform in chronic care would save time and effort to have it successfully implemented.

Similar to the U.S., health policy makers in Europe considered also the structure of the financial incentives of key stakeholders in each country when designing payment

schemes for integrated care. As a result, newly introduced payment schemes in Europe targeted those stakeholders who were expected to adjust their behaviour and provided them with adequate financial incentives. This is crucial factor of a successful reform considering that misaligned incentives between health care stakeholders were found to be the strongest barriers in payment reform. However, the consecutive reforms denote that the right mixture and level of financial incentives to stimulate stakeholders towards integrated care is still to be found in Europe.

A solution to this would ideally be the integration of payment of all necessary health services for people with chronic diseases. Part A in this thesis showed that the implemented payment schemes to integrated care in European countries achieved different targets. For example, PFC appeared to be the most successful in increasing collaboration within and across health care sectors, whereas bundled payment was perceived to integrate financing of different care sectors. In addition, the implemented payment schemes appeared able to reduce the growth of health care expenditure in different health care sectors. For example PFC and all-inclusive payments reduced the growth in outpatient expenditure while PFP reduced the growth in hospital expenditure. Therefore, a blended payment scheme that has a yearly risk-adjusted global payment as basis and PFC and PFP as additional payments is likely to overcome the barriers of each individual scheme and provide the strongest financial incentives to control health care expenditure. Similar blended payment schemes in the U.S. proved to improve quality of care and reduce health care expenditure [46].

The designers of such a blended payment structure would face a number of challenges. First, it should be thoroughly considered whether the global payment to health care providers would be per insured, per patient, per citizen or per specific population. The structure of a health care system should be included in this consideration. For example, a global payment per specific population would be easier to be implemented in U.K. where NHS is the main purchaser of care than in The Netherlands were several health insurers purchase care within a region. Second, the D&I costs of DMPs (when applicable) should also be incorporated in a blended payment. These costs are expected to fluctuate over time due to economies of scale, economies of scope and the cumulative management experience. This fluctuation would be difficult to be incorporated in the (negotiated) global payment component. Third, the PFP component should reward good performance on various outcomes of integrated care including indicators related to multi-morbidity, prevention and self-management. Fourth, the global payment and the PFP components of a blended scheme should be designed in a way that discourages health care providers to "game" (by "upcoding" the severity of the patients and aiming only to score high on the performance indicators instead of improving the quality of care, respectively) the payment system.

As Part A showed, "gaming" of payment schemes and misaligned incentives of stakeholders were the strongest barriers of payment reforms. These barriers may also be present in the introduction of a blended payment scheme as described above. Shared savings could also be added as additional payment to the blended payment scheme in order to overcome these barriers. Under this payment component, one or more care providers and one or more health insurers form an agreement that savings relative to a benchmark can be returned to the providers and/or insurers. Therefore, health care providers and payers have aligned incentives to contain costs and improve care quality. To achieve full alignment across stakeholders, the allocation of gains among the participating health care providers should financially satisfy all involved parties [261]. Furthermore, a part of the gains could be used to support intensive prevention and self-management support programs and reward patients (e.g. by reducing co-payments for patients who achieved their healthy behaviour goals).

The last and probably the most important ingredient in designing and introducing an appropriate payment scheme for integrated care, is the strong willingness and commitment of the health care authorities to facilitate this process. Re-positioning financial incentives and changing behaviour in health care sector, which requires a large share of a country's GDP and workforce, is not an easy task.

10.3 Determinants of DMP cost-effectiveness

The evidence about the cost-effectiveness of DMPs is inconclusive [2,49]. A reason for this is that the nature of a DMP differs substantially from the conventional medical technologies (e.g. drugs, devices, treatment equipment, surgical interventions, screening, and vaccination programs) that have most commonly been subjected to economic evaluation. DMPs commonly contain a mixture of many different interventions that involve changes in the process of the chronic care delivery and the behaviour of caregivers and patients. Part B and Part C in this thesis showed that the cost-effectiveness of DMPs vary considerably and it is most likely depending on the components and intensity of the program, the success of the implementation, the (case-mix of the) target population, the D&I costs, and time.

On the effectiveness side, DMPs that provided a larger number of interventions across the six components of the CCM were found to be potentially cost-saving, effective or cost-effective (see Chapter 8). However, the effectiveness depends also on the mixture of interventions provided by a DMP. Different combinations of interventions within and across the components of the CCM could lead to different effects of the program. The results in Chapter 7 indicated that interventions that stimulate physical activity and self-efficacy were predictive of an increase in patient's quality of life. Therefore, the inclusion

of these interventions in a DMP would increase its effectiveness. Furthermore, patients with multiple morbidities seem to benefit less than patients with one disease. This may imply that the current disease-specific DMPs do not address the needs of patients with multi-morbidity and therefore, they are less effective for this population.

It is also expected that the effectiveness of DMPs would be higher, if the intensity level of the interventions would be carefully determined and customised to the needs of the participating patients. An appropriate mixture of interventions but at low intensity level would presumably not be effective to chronic patients with high unmet needs.

Moreover, Part C in this thesis reported that comprehensive DMPs seem to be more appropriate to address the needs of more severe or complex patients. This means that the comprehensiveness of the DMPs should be aligned with the case-mix of patients to address effectively the needs of patients especially of those with multi-morbidity. In addition, the motivation and health literacy of the target population determines the effectiveness of the DMP [12]. Patients that are not willing or not able to become health managers and comply with treatment plans would barely get benefits from DMPs, thereby limiting its effectiveness on the aggregated level.

It is obvious that if the provided interventions are not successfully implemented then the effectiveness (if any) of the DMP would be limited. Therefore, finding the right mixture and density of interventions that is optimally comprehensive for a specific target population and successfully implemented is the recipe to maximize the effectiveness of DMPs. Successful implementation strategy requires carefully planning that takes into account the nature of the innovation; characteristics of the professionals and patients involved; and the social, organisational, economic and political context [262].

On the cost side, Part B of this thesis reported wide variation in D&I costs of DMPs, which is driven primarily by the duration of the development phase, the labor intensiveness needed to develop and implement a DMP, and the size of the providing organization. As a result, the D&I costs would be lower in DMPs with experienced and capable DMP managers, developed infrastructure (e.g. ICT system), and high level of care integration prior to the development phase. These costs would also be lower if existing economies of scale and economies of scope would be optimally explored and exploited. This means that once an organization has invested in developing and providing a DMP, it should utilize the accumulated knowledge and infrastructure to include more patients in the DMP and to provide more DMPs. By doing this, the costs of providing a DMP are expected to become lower, thereby increasing its cost-effectiveness.

Furthermore, it should be realized that if the D&I costs outweigh the potential cost savings of a DMP then its cost-effectiveness is at stake. The sensitivity analysis in Chapter 7 and Chapter 8 showed that the inclusion of D&I costs did not change the cost-effectiveness of the compared DMPs. However, it should be reminded that the analysis

compared DMPs. If a DMP was compared to usual care, where there are no D&I costs, the impact of these costs on the cost-effectiveness of a DMP would be larger.

Time is undoubtedly a major determinant of the cost-effectiveness of DMPs. It is expected that evident improvements in quality of care and patient health behavior would slow down disease progression, reduce symptoms, prevent events (e.g. stroke) and avoid complications (e.g. diabetic foot ulcers). This would increase the cost-effectiveness of DMPs in the long term by improving patient's quality of life (or even mortality) and reducing hospitalization costs, which are the main drivers of total DMP costs.

10.4 Relevance and implications for stakeholders in the Dutch chronic care

Chapter 3 highlighted possible threats to and necessary conditions for the success of the bundled payment in the Netherlands. Almost five years after the introduction of this payment scheme, these conditions have not been met and the possible threats are still present [93,196]. Five major conditions remain unfulfilled in the implementation of bundled payment. These include the adequate definition and measurement of performance indicators, the well-performing integrated ICT systems, the alignment of the provided DMP interventions and the targeted population, and the inclusion of a greater variety of health services in the bundled payment.

The current Dutch performance indicators are based on clinical guidelines and protocols and focus more on the process of care delivery rather than on outcome measurements. However, the current set of performance indicators does not include measures related to the provision of patient-tailored care, adequate treatment of multimorbid patients, and coordination of multi-disciplinary teams. These processes are core elements in the provision of integrated care and improvements in them should be measured and rewarded. Further, performance indicators targeting disadvantaged groups and economically deprived population should be included in the reward of health care providers. A study showed that tackling health inequalities has a larger effect on patient health than the current performance indicators included in the British Quality and Outcomes Framework [260]. Chapter 2 showed a general trend of PFP schemes towards the inclusion of process performance indicators that adequately reflect the aims of the payment scheme. This is because process performance measurements capture important information that is otherwise not measured and may have large effects on patient health [263]. In addition, the performance is currently measured at the organizational-level and it does not reflect the needs of a specific population. Patient-specific or group-specific (e.g. patients with multi-morbidity) performance indicators tailored to the needs of a specific population group would enable the reward of patient-tailored care. An example

would be to reward health care providers for the holistic assessment of patients with multi-morbidity and for monitoring the interaction of their medical treatment for different diseases. Another example would be that reducing body mass index among obese patients should be rewarded regardless the fact that they are still considered as overweight (i.e. their BMI is above a specific threshold). Measuring both absolute and relative improvements on specific indicators would stimulate high-performing providers to maintain their performance levels and motivate low-performing providers to achieve relatively high performance [264].

The last five years, there were many attempts to establish effective communication between ICT systems (e.g. between primary and secondary care), implement integrated ICT systems, and register clinical data appropriately in the ICT systems. Remarkable improvements have been made ever since. However, the result of these attempts is far from having achieved well-performing ICT systems able to facilitate integration of chronic care. Despite recent improvements, the patient data is still not adequately registered and sufficiently shared between different health care providers. Chapter 6 indicated that an ICT system is a driver of D&I costs and suggested the collective involvement and support of health insurers, care organizations and authorities in their full development. The recognition of the shared benefits of an ICT development by the stakeholders may be not enough to trigger their involvement and active participation. Practical solutions to the technical and privacy challenges of linking ICT systems should also be provided. In addition, providers would be (more) motivated to register processes and outcomes if these registration data were used in the clinical practice (e.g. to guide treatment choices), public reporting, and/or to support payment schemes.

Furthermore, Chapter 5 pointed out that DMPs included sub-group of patients with high costs for which the potential cost savings due to DMPs may be higher than for other patients and Chapter 7 concluded that patients with multi-morbidity seem to benefit less from DMPs than patients with a single chronic disease. The DMPs, as currently implemented in the Netherlands, provide the same set of interventions to patients with similar clinical profiles without sufficient incorporation of patient needs, preferences and capabilities. Taking into account the variability in patient needs with one or more chronic diseases, this approach of providing DMPs may be not efficient. Predictive modelling to identify a group of patients that can potentially obtain the maximum effectiveness of a specific set of DMP interventions should be applied during the development phase of DMPs. For example, Chapter 7 showed that the inclusion of physical activity interventions in COPD-DMPs and the support of COPD patients to get or remain in employment are linked with reduced health care costs of these patients. In this approach, several sets of DMP interventions could be combined to provide a patient-tailored package of disease management interventions. This is also discussed in Chapter 8 where more comprehensive DMPs appeared to address more adequately

the needs of severe/complex patients than less comprehensive DMPs. Moreover, it is evident that the cost-effectiveness of DMPs would increase by providing customized set of interventions to chronic patients. This would trigger health insurers to invest in such programs, which is a condition for the success of the bundled payment (Chapter 3). At the same time, the financial risk exposure of the DMP providers would be reduced if the negotiated bundled payment would adequately differentiate (i.e. full risk equalization) based on the case-mix of patients that receive a specific set of interventions and would fully include the DMP development and implementation costs.

The bundled payment is disease specific and includes only a part of the total health care expenditure for chronic care (e.g. physiotherapy, smoking cessation support, hospital care and medication are not or to fully included in this payment). The narrow scope of services included in this payment enfolds two threats for the success of the scheme from the list of threats described in Chapter 3. There is a risk of segmentation of care between a) contracted and non-contracted providers, b) different diseases per patient, and c) services covered in the basic and supplementary health insurance packages. Further, bundled payment and registration of performance indicators improved the transparency in health service delivery but double payments and other inefficiencies are not eliminated. This withholds the alignment of the financial incentives between health care stakeholders which is an important facilitator of the success of a payment scheme (see Chapter 2). To overcome the narrow scope of bundled payment, global payment as implemented in the U.S. would be necessary to be in place. One of the most popular models of global payment is the alternative quality contract (AQC) implemented in the BlueCross Blue Shield of Massachusetts, which combines a health status–adjusted global payment with performance incentives for meeting quality and safety benchmarks. Contracted providers can improve margins through quality bonuses of up to 10 percent and by reducing spending growth below the level of inflation [149]. However, such a payment would require adequate performance measurement with data from well-developed ICT systems for a set of interventions tailored to patient needs, as described above. These elements are included in the current initiatives towards the so called "population based budgeting" which is a global payment adapted to the Dutch health care setting. Hopefully the findings of this thesis about payment schemes for integrated care and their barriers for success as well as the potentials of providing DMPs for improving outcomes and reducing costs will be considered in the design of the next Dutch payment scheme for integrated care.

10.5 Methodological challenges and responses in the evaluation of integrated care

As stated in the literature, the economic evaluation of DMPs is challenging [49], a fact that is reflected in large variation in objectives, designs, indicators, target audiences, and actors involved [206] and the inconclusive evidence [40].

The first challenge includes the definition of DMPs (or integrated care programs) because there is a diversity of interventions and intervention intensity provided by these programs. Even the same set of interventions would have been differently implemented in other settings and countries. This variation in the definition of DMPs results in treatment variations. The specific interventions and their intensity as well as the specific context (i.e. targeted patient population, organizational experience with integrated care, relevant policies at national level) should be recorded and stated in order to provide an understanding of the "treatment". A detailed list (as used in Chapter 8 and Chapter 9) with implemented interventions completed and updated periodically by the providing organization complemented with qualitative data about the context of the intervention could ensure the sufficient definition of an evaluated DMP.

Further, establishing the counterfactual of DMPs is difficult. Identifying health care providers and patients willing to take the chance to be assigned to a control group that does not participate in a DMP is difficult, even if one thinks that it is still ethically acceptable. Further, DMPs often include interventions at organizational-level such as redesign of care delivery and new ICT systems. These interventions are likely to affect all patients within an organization increasing therefore, the risk of contamination from an intervention group (patients participating in a DMP) to a control group (patient not participating in a DMP). Thus, it is difficult to identify a control group of patients within an organization which provides DMP. This can be avoided by adopting a cluster-RCT were the organizations (not patients) are randomly allocated to the treatment and control groups. However, such a design requires substantial financial resources and commitment from many health care providers in different organizations. Randomization may be also difficult to be achieved as researcher may have limited power in determining the participating organizations in a DMP. Even if a control group is adequately specified, national or regional quality improvement policies and initiatives may influence the treatment in the control group. To overcome this, a detailed list of the interventions offered to the control group should be recorded similar to the intervention group. As a result, many evaluation studies of DMPs adopted observational study design. Chapter 7 and Chapter 8 demonstrated retrospective statistical methods such as propensity score matching that can be used to reduce confounding from non-randomisation. However, an optimal propensity score matching would require a relatively large sample size which is often not the case in the evaluation of DMPs. In addition, DMP evaluations are

frequently subject to selection bias because a specific group of patients tends to be included in DMPs (e.g. those motivated to change, those with frequent disease-episodes or those with a higher SES). Nevertheless, the response rate is frequently problematic. For the latter, missing data analysis and multiple imputation as performed in Chapter 5 or repeated measurement analysis as performed in Chapter 8 could be used to reduce selection bias. However, these techniques are most suitable when the percentage of missing observations is not too large [185,192]. Repeated measurement analysis (i.e. analysing of longitudinal data) could also reduce biases due to regression to the mean.

Another challenge is the selection of indicators and instruments to measure outcomes and costs. DMPs aim to improve the quality of the care process, improve patients' self-management abilities, enhance patients' health and reduce costs. Thus, different measures should be employed in the evaluation which makes the data collection process laborious for researchers (demanding in financial resources and time), patients (lengthy questionnaires at the expense of the response rate), and health care providers (lengthy questionnaires and cumbersome data extraction from ICT systems). As a result the data richness and availability is at stake. Carefully selecting validated measures that are expected to capture the effect of DMPs on key outcomes (e.g. biochemically validated smoking rates when smoking cessation interventions are included in the DMP and disease-specific quality of life instruments) may overcome this challenge.

Another challenge is the determination of an appropriate follow-up period in the evaluation of DMPs. This thesis argued that improvements in process and intermediate outcomes take a long time to be reflected in final outcomes. This would be solved by adopting long follow-up periods in the evaluation but that would discourage patients and health care providers to participate in the evaluation and increase the required research budget. It would also increase the uncertainty of the results because the care delivered in DMPs (and eventually in control groups) would be more likely to have changed due to updates of clinical guidelines and further health care reforms. Hence, using observational data to estimate long-term effects is unavoidable.

10.6 Recommendations for future research

George Bernard Shaw once said in a toast to Albert Einstein "Science is always wrong, it never solves a problem without creating ten more." This thesis is not an exception to this argument. While it provided evidence about the payment and economic evaluation of integrated care, it also highlighted numerous challenges to be addressed in future research.

With regards to the payment of integrated care, future research may investigate the impact of financial agreements on a wide range of outcomes (e.g. quality of care

and morbidity) in the long-term using a large dataset at national/regional/organizational level with more observations after implementation of a payment scheme. In PFP schemes, future research may determine the optimal level of payment to accelerate quality improvement and investigate net cost savings (i.e. financial rewards subtracted from costs savings in health care utilization). Such research is data-intensive, requiring time-series on targeted and other outcome measures for intervention and (credible) control group.

Regarding the variability in costs of DMPs, further research can investigate whether a broader set of factors (than those included in Chapter 5) has a relation with health care costs of patients included in DMPs. Such factor could include, on DMP-level, the size of a DMP, the number and type of care providers involved in a DMP and the intensity (i.e. frequency and type of interventions) of DMPs. On patient-level, they may include more indicators of disease severity, prior health care utilization, health literacy, disease perceptions and self-management behaviour. An investigation of cost patterns in patients with and without multi-morbidity focusing on the combination of morbidities could also be an interesting topic for further investigation. In addition, future research should investigate in details the existence of economies of scale and economies of scope in the provision of DMPs and their relation with the D&I costs of DMPs.

With respect to the evaluation of DMPs, future research may use predictive modelling (as suggested in Chapter 7) to determine patient profiles for specific sets of DMP interventions as well as to investigate how the comprehensiveness (expressed in number of interventions and intensity) and the case-mix of patients determines the cost-effectiveness of these programs (as suggested in Chapter 8). Finally, future research may identify a core or minimal set of criteria and indicators for each objective to be used in the evaluation framework presented in Chapter 9 and obtain weights for each criterion that reflect stakeholder preferences, to enable multi-criteria decision analyses.

CHAPTER 11

Summary and author's information

11.1 Summary

Chronic diseases have an increasingly negative impact on a) population health by increasing morbidity and mortality, b) society by increasing health inequalities and burden to informal care givers, and c) economy by requiring enormous financial resources and jeopardizing macro-economic development (e.g. consumption, capital accumulation, labour productivity and labour supply). Integrated care is the most promising concept in redesigning care to tackle the increasing threat of chronic diseases. Several European countries have experimented with models for integrating care, most frequently in the form of disease management programs (DMPs). These models were often supported by payment schemes to provide financial incentives to health care providers for implementing integrated care. This thesis aimed to investigate these payment schemes and assess their impact (Part A), explore the variability in costs of DMPs (Part B), and determine the costs and effects of DMPs (Part C).

Part A- Payment of integrated care

Chapter 2, explored the adoption and success of payment schemes that promote integration of chronic care in European countries. A literature review and fifteen interviews with experts in care for chronic diseases were carried out to obtain detailed information regarding the payment schemes, facilitators and barriers to their implementation, and their perceived success. Austria, France, England, the Netherlands, and Germany have implemented payment schemes that were specifically designed to promote the integration of care for chronic patients. Prominent factors facilitating implementation included stakeholder cooperation, adequate financial incentives for stakeholders, and flexible task allocation among different care provider disciplines. Commonly reported barriers to implementation included misaligned incentives across stakeholders and gaming. The implemented payment schemes targeted different stakeholders in different countries depending on the structure and financing of each health care system. All payment reforms appeared to have changed the structure of care for chronic patients. Pay-for-coordination (PFC), as it was implemented in Austria, France and Germany, was perceived to be the most successful in increasing collaboration within and across health care sectors, whereas pay-for-performance (PFP), as it was implemented in England and France, was perceived most successful in improving other indicators of the quality of the care process such as coordination in primary care. The interviewees stated that the impact of the payment reforms on health care expenditures remained questionable. The chapter concluded that the success of a payment scheme depends on the details of the specific implementation in a particular country, but a combination of the schemes may overcome the perverse incentives in each individual scheme.

Chapter 3, described the recent attempts in the Netherlands to stimulate the delivery of integrated care for chronic patients, focusing specifically on the new integrated payment scheme and the barriers to introducing this scheme. Based on literature, government documents, personal communications and site visits to DMPs, the most important conditions for the success of the new payment scheme were identified including complete care protocols describing both general and chronic disease-specific care modules, coverage of all components of a DMP by basic health care insurance, adequate information systems that facilitate communication between care givers, explicit links between the quality and the price of a DMP, expansion of the amount of specialized care included in the chain-DTC (i.e. bundled payment), inclusion of a multi-morbidity factor in the risk equalization formula of insurers, and thorough economic evaluation of DMPs.

Chapter 4 studied the impact of financial agreements, which were introduced in European countries to stimulate integration of care, on health care expenditure. This study used difference-in-differences models to estimate differences in health care expenditure trends before and after the introduction of the payment reforms using OECD and WHO data from 1996 to 2013 from 9 intervention countries and 16 control countries. Intervention countries included countries with PFC, PFP, and/or all-inclusive agreements (bundled and global payment) for integrated care. The results showed that the annual growth of outpatient expenditure was decreased in countries with PFC (by 21.28 US$ per capita) and in countries with all-inclusive agreements (by 216.60 US$ per capita) at the year of implementation. The growth of hospital and administrative expenditure was decreased in countries with PFP by 64.50 US$ per capita and 5.74 US$ per capita, respectively. During the total 4-year period after implementation, PFP decreased the growth of hospital and administrative expenditure and all-inclusive agreements reduced the growth of outpatient expenditure. The chapter concluded that financial agreements are potentially powerful tools to stimulate integrated care and influence health care expenditure. A blended payment scheme that combines elements of PFC, PFP, and all-inclusive payments is likely to provide the strongest financial incentives to control health care expenditure.

Part B- Cost variation in disease management programs

Chapter 5 identified factors on patient-level and organizational-level that explain the variability in costs of patients with different chronic diseases enrolled in a DMP. Generalized linear mixed models were specified to perform a multi-level analysis of cross-sectional data from 16 DMPs across 3 disease areas in the Netherlands. Multiple imputation, subgroup analysis per disease and analysis from both the health care and the societal perspectives were also performed. The results showed that age, the presence of cardiovascular disease, multi-morbidity and additional payments on top of the base payment for the usual care had a positive relation with costs, while better quality

of life was associated with lower health care costs. In the COPD sample, physical activity and employment were associated with lower health care costs. This chapter showed that there is great variability in health care costs among patients included in DMPs and identified patient and organizational explanatory factors. The findings are relevant to the design of future DMPs and their payment schemes.

Chapter 6 investigated the variability in and drivers of D&I costs among 22 Dutch DMPs and highlighted characteristics that impact them by using descriptive statistical analysis, document analysis and interviews. The development costs varied from €5,891 to €274,783 and the implementation costs varied from €7,278 to €387,879 across DMPs. Personnel costs were the main component of development costs. Development costs were strongly correlated with the implementation costs ($\rho = 0.55$), development duration ($\rho = 0.74$), and number of FTEs dedicated to DMP development. Organizations with a large size and high level of integrated care prior to the implementation of a DMP had relatively low development costs. These findings were in line with the cross-case qualitative comparison where programs with a longer history, more experienced project leadership, previously established ICT systems, and less complex patient populations had lower D&I costs. The chapter concluded that there is wide variation in D&I costs of DMPs, which is driven primarily by the duration of the development phase and the staff needed to develop and implement a DMP. These drivers are influenced by the attributes of the DMP, characteristics of the target population, project leadership, and ICT involved. There are indications of economies of scale and economies of scope, which may reduce D&I costs.

Part C- Economic evaluation of integrated care

Chapter 7 investigated the 1-year changes in costs and effects of 1,322 patients in 16 DMPs for cardiovascular risk (CVR), chronic obstructive pulmonary disease (COPD), and diabetes mellitus (DMII) in the Netherlands and explored the within-DMP predictors of these changes. A cost-utility analysis was also performed from the health care and societal perspective comparing the most and the least effective DMP within each disease category. The results showed wide variation in development and implementation costs between DMPs (range: €16;€1,709) and highlighted the importance of economies of scale. Changes in health care utilization costs were not statistically significant. DMPs were associated with improvements in integration of CVR care (0.10 PACIC units), physical activity (+0.34 week-days in which patients had at least 30 minutes of moderate or severe physical activity) and smoking cessation (8% less smokers) in all diseases. Since an increase in physical activity and in self-efficacy were predictive of an improvement in quality-of-life, DMPs that aim to improve these are more likely to be effective. When comparing the most with the least effective DMP in a disease category as defined by the EQ5D, the vast majority of bootstrap replications (range:73%;97%) pointed to cost

savings, except for COPD (21%). QALY gains were small (range:0.003;+0.013) and surrounded by great uncertainty. The chapter concluded that after one year there were indications of improvements in level of integrated care for CVR patients and lifestyle indicators for all diseases, but in none of the diseases we have found statistically significant cost savings due to DMPs. However, it is likely that it takes more time before the improvements in care lead to reductions in complications and hospitalizations and hence to cost savings.

Chapter 8 compared the physical activity, smoking status, quality-adjusted life years (QALYs) and yearly costs per patient between the most and the least comprehensive DMP in four disease categories: primary CVR-prevention, secondary CVR-prevention, both types of CVR-prevention, and COPD (n:1,034). Propensity score matching increased comparability between DMPs. A two-year cost-utility analysis (CUA) was performed from the health care and societal perspective. Sensitivity analysis was performed to estimate the impact of DMP development and implementation costs on the cost-effectiveness. The results showed that patients in the most comprehensive DMPs increased their physical activity more (except for the primary CVR-prevention) and had higher smoking cessation rates. The incremental QALYs ranged from -0.032 to 0.038 across all diseases. From a societal perspective, the most comprehensive DMP decreased costs in the primary CVR-prevention (certainty: 57%), secondary CVR-prevention (certainty: 88%), and both types of CVR-prevention (certainty: 98%). Moreover, the implementation of comprehensive DMPs led to QALY gains in secondary CVR-prevention (certainty: 92%) and COPD (certainty: 69%). The chapter concluded that the most comprehensive DMPs for CVR and COPD have the potential to be cost-saving, effective, or cost-effective compared to the least comprehensive DMPs. The challenge for Dutch stakeholders is to find the optimal mixture of interventions that is most suited for each target group.

Chapter 9, developed a methodological framework to facilitate the application of Multi-Criteria Decision Analysis (MCDA) for a comprehensive economic evaluation of disease management programs (DMPs). This was achieved by studying previously developed frameworks for the evaluation of DMPs and different MCDA methods and using practical field experience in the economic evaluation of DMPs and personal discussions with stakeholders in chronic care. The framework includes different objectives and criteria that are relevant for the evaluation of DMPs, indicators that can be used to measure how DMPs perform on these criteria, and distinguishes between the development and implementation phase of DMPs. The objectives of DMPs are categorised into a) changes in the process of care delivery, b) changes in patient lifestyle and self-management behaviour, c) changes in biomedical, physiological and clinical health outcomes, d) changes in health-related quality of life, and e) changes in final health outcomes. All relevant costs of DMPs are also included in the framework. Based on this framework we conducted a MCDA of a hypothetical DMP versus usual care. The chapter calls for a com-

prehensive economic evaluation of DMPs that is not just based on a single criterion but takes into account multiple relevant criteria simultaneously. The framework presented in this chapter is a step towards standardising such an evaluation.

Chapter 10, provided a general discussion starting with the main findings of the preceding chapters. In this chapter, a blended payment scheme was suggested to overcome potential barriers to the successful payment reform by providing an appropriate mix of financial incentives to health care providers and health payers. The structure of a health care system should be considered when designing such a blended payment scheme and the DMP development and implementation costs should be incorporated in the payment. Further, the PFP component should reward good performance and a shared savings scheme could be used to avoid "gaming" and align the incentives of the stakeholders. Moreover, this chapter argued that finding the right mixture and density of DMP interventions that is optimally comprehensive for a specific target population and successfully implemented is the recipe to maximize the effectiveness of DMPs. The implications of the thesis' findings were also discussed in Chapter 10. They were mainly related to the adequate definition and use of performance indicators, the design of a well-established ICT integrated system, the alignment of the DMP interventions with the targeted population, and the greater inclusion of health services in the bundled payment. The methodological challenges and potential solutions to overcome them as well as suggestions for future research conclude Chapter 10.

11.2 Samenvatting

Chronische ziekten hebben een steeds grotere weerslag op a) de volksgezondheid: stijgende morbiditeit en mortaliteit, b) de samenleving: toenemende ongelijkheid in gezondheid en een groter beroep op informele zorgverleners, en c) de economie: een groter beslag op de financiële middelen dat de macro-economische ontwikkelingen in gevaar brengt (bijv. consumptie, kapitaalaangroei, arbeidsproductiviteit en aanbod op de arbeidsmarkt). Als het gaat om een herontwerp van de zorg met als doel de toenemende dreiging van chronische ziekten aan te pakken, is integrale zorg het meest veelbelovende concept. In verschillende Europese landen wordt geëxperimenteerd met modellen voor integrale zorg, meestal in de vorm van 'disease management programma's' (DMP's). Deze modellen worden vaak ondersteund door een bekostigingssystematiek die zorgaanbieders een financiële prikkel moeten bieden om integrale zorg te implementeren. Dit proefschrift heeft als doel deze bekostigingssystemen te onderzoeken en het effect ervan te beoordelen (deel A), de variabiliteit van de kosten van DMP's te verkennen (deel B) en vast te stellen welke kosten en effecten DMP's met zich meebrengen (deel C).

Deel A- De bekostiging van integrale zorg

Hoofdstuk 2 biedt een verkenning van de invoering en het succes van bekostigingssystemen ter bevordering van de integratie van zorg voor chronisch zieken in Europese landen. Er is een literatuuronderzoek uitgevoerd en er zijn vijftien interviews gehouden met deskundigen op het gebied van de zorg voor chronisch zieken teneinde uitvoerige informatie te verkrijgen over de bekostigingssystemen, de factoren die de implementatie van deze systemen bevorderen en belemmeren en het veronderstelde succes ervan. Oostenrijk, Frankrijk, Engeland, Nederland en Duitsland hebben bekostigingssystemen ingevoerd die specifiek gericht waren op het bevorderen van de integratie van de zorg voor chronisch zieken. Belangrijke factoren die de implementatie bevorderden waren samenwerking tussen belanghebbenden, adequate financiële prikkels voor belanghebbenden en een flexibele taakverdeling over verschillende disciplines van zorgaanbieders. Vaak genoemde belemmeringen voor de implementatie waren verkeerd afgestemde prikkels voor belanghebbenden en speculatief gedrag. De bekostigingssystemen waren in verschillende landen gericht op verschillende belanghebbenden, afhankelijk van de structuur en financiering van het betreffende zorgstelsel. Alle bekostigingshervormingen bleken te hebben bijgedragen aan een verandering van de structuur van de zorg voor chronische patiënten. PFC (pay-for-coordination), een systeem waarbij expliciet wordt betaald voor de coördinatie van zorg, zoals dat in Oostenrijk, Frankrijk en Duitsland werd ingevoerd, werd ervaren als de meest succesvolle manier om de samenwerking binnen en tussen gezondheidszorgsectoren te verbeteren. PFP (pay-for-performance), zoals dat

in Engeland en Frankrijk werd ingevoerd, werd ervaren als de meest succesvolle manier om andere indicatoren van de kwaliteit van het zorgproces, zoals de coördinatie van de primaire zorg, te verbeteren. De geïnterviewden gaven aan dat het effect van de bekostigingshervormingen op de uitgaven voor de gezondheidszorg twijfelachtig was. De conclusie van het hoofdstuk is dat het succes van een bekostigingssysteem afhangt van de bijzonderheden van de specifieke implementatie in een bepaald land, maar dat de perverse prikkels in ieder afzonderlijk systeem door een combinatie van verschillende systemen kan worden ondervangen.

Hoofdstuk 3 bevat een beschrijving van de recente pogingen in Nederland om de levering van integrale zorg voor chronische patiënten te bevorderen, met specifieke aandacht voor de nieuwe integrale bekostigingssystematiek en de belemmeringen voor de invoering daarvan. Op basis van literatuur, overheidsdocumenten, persoonlijke communicaties en bezoeken ter plaatse aan DMP's zijn de belangrijkste voorwaarden voor het welslagen van de nieuwe bekostigingsregeling in kaart gebracht, waaronder protocollen voor complete zorgtrajecten waarin zowel algemene zorgmodulen als zorgmodulen specifiek gericht op bepaalde chronische ziekten worden beschreven, dekking van alle onderdelen van een DMP door de basisverzekering voor zorgkosten, toereikende informatiesystemen die de communicatie tussen zorgverleners vereenvoudigen, expliciete verbanden tussen de kwaliteit en de prijs van een DMP, uitbreiding van de hoeveelheid specialistische zorg die is opgenomen in de keten-DBC (d.w.z. gebundelde betaling), opname van een multimorbiditeitsfactor in het risicovereveningsmodel van verzekeraars en een uitvoerige economische evaluatie van DMP's.

In **hoofdstuk 4** wordt gekeken naar het effect van financiële overeenkomsten die in Europese landen zijn uitgevoerd om de integratie van zorg te bevorderen, op uitgaven voor de gezondheidszorg. Voor dit onderzoek is gebruikgemaakt van "difference-in-differences"-modellen om een inschatting te maken van verschillen in trends in de uitgaven voor de gezondheidszorg vóór en na de invoering van de bekostigingshervormingen op basis van gegevens van de OECD en de WHO van 1996 tot 2013 uit 9 interventielanden en 16 controlelanden. De interventielanden waren landen met PFC, PFP, en/of alomvattende bekostigingssystemen (gebundelde bekostiging en alomvattende budgetten) voor integrale zorg. De resultaten wezen uit dat de jaarlijkse stijging van de uitgaven voor poliklinische zorg in landen met PFC en landen met alomvattende bekostigingssystemen in het jaar van invoering was afgenomen met respectievelijk 21,28 dollar per capita en 216,60 dollar per capita. De stijging van de ziekenhuiskosten en administratieve uitgaven was in landen met PFP afgenomen met respectievelijk 64,50 dollar per capita en 5,74 dollar per capita. Gedurende de gehele periode van vier jaar na invoering leidde PFP tot een afname van de stijging van ziekenhuiskosten en administratieve uitgaven en resulteerde alomvattende bekostiging in een afname van de stijging van de uitgaven voor poliklinische zorg. De conclusie van het hoofdstuk is

dat financiële overeenkomsten potentieel krachtige instrumenten zijn om integrale zorg te bevorderen en de uitgaven voor de gezondheidszorg te beïnvloeden. Een gemengd bekostigingssysteem waarin elementen van PFC, PFP en alomvattende bekostiging worden gecombineerd, zal naar verwachting de krachtigste financiële prikkels ter beheersing van de uitgaven voor de gezondheidszorg bieden.

Deel B- Variatie in de kosten van programma's voor ziektebeheer

In **hoofdstuk 5** worden factoren op patiëntniveau en organisatieniveau aangewezen waarmee de variabiliteit van de kosten van patiënten met verschillende chronische ziekten die aan een DMP deelnemen, kunnen worden verklaard. Er zijn gegeneraliseerde lineaire gemengde modellen gebruikt om multi-level analyses van cross-sectionele data van de 16 DMP's verspreid over 3 ziektegebieden in Nederland uit te voeren. Ook zijn meervoudige imputaties, subgroepanalyses per ziekte en analyses vanuit zowel het gezondheidszorgperspectief als het maatschappelijk perspectief uitgevoerd. De resultaten wezen uit dat er een positieve relatie bestaat tussen leeftijd, de aanwezigheid van cardiovasculaire ziekte, multimorbiditeit en aanvullende bekostiging boven op de basisbekostiging voor de gebruikelijke zorg enerzijds en de kosten anderzijds. Een betere kwaliteit van leven was geassocieerd met lagere kosten van de gezondheidszorg. In de COPD-groep waren lichaamsbeweging en het hebben van betaald werd geassocieerd met lagere kosten van de gezondheidszorg. Dit hoofdstuk toont aan dat er sprake is van grote variabiliteit in de kosten van de gezondheidszorg onder patiënten die aan DMP's deelnemen en dat die variabiliteit samenhangt met zowel patiëntkenmerken als organisatiekenmerken. De bevindingen zijn relevant voor de opzet van toekomstige DMP's en de daarbij behorende bekostigingssystematiek.

In **hoofdstuk 6** wordt de variabiliteit van de kosten voor ontwikkeling en implementatie van 22 Nederlandse DMP's onderzocht, alsmede de factoren die deze kosten bepalen. Dit gebeurt aan de hand van een descriptieve statistische analyse, een documentanalyse en interviews. De ontwikkelingskosten van de verschillende DMP's varieerden van € 5.891 tot € 274.783 en de implementatiekosten van € 7.278 tot € 387.879. De personeelskosten vormden het belangrijkste onderdeel van de ontwikkelingskosten. De ontwikkelingskosten hingen nauw samen met de uitvoeringskosten ($\rho = 0{,}55$), ontwikkelingsduur ($\rho = 0{,}74$) en het aantal FTE's dat werd ingezet voor de ontwikkeling van DMP's. Bij grote organisaties die voorafgaand aan de implementatie van een DMP een hoog niveau van integrale zorg kenden, waren de ontwikkelingskosten relatief laag. Deze bevindingen waren in overeenstemming met de kwalitatieve vergelijking tussen programma's, waarbij programma's met een langere geschiedenis, projectleiders met meer ervaring, eerder ingevoerde ICT-systemen en minder complexe patiëntenpopulaties lagere kosten voor ontwikkeling en implementatie hadden. De conclusie van dit hoofdstuk is dat de kosten voor de ontwikkeling en implementatie van DMP's sterk

uiteenlopen, waarbij de duur van de ontwikkelingsfase en het personeel dat nodig is om een DMP te ontwikkelen en te implementeren, de voornaamste bepalende factoren zijn. Deze factoren worden beïnvloed door de kenmerken van het DMP, kenmerken van de doelpopulatie, projectleiding en de hierbij betrokken ICT. Er zijn aanwijzingen dat er sprake is van schaalvoordelen en toepassingsvoordelen, die tot lagere kosten voor ontwikkeling en implementatie kunnen leiden.

Deel C- Economische evaluatie van integrale zorg

Hoofdstuk 7 bevat een beschrijving van een onderzoek naar de veranderingen in de kosten en effecten gedurende één jaar bij 1.322 patiënten in 16 DMP's voor cardiovasculair risico (CVR), chronische obstructieve longziekte (COPD) en diabetes mellitus (DMII) in Nederland. Ook worden in dit hoofdstuk de predictoren van deze veranderingen verkend. Er werd ook een kostenutiliteitsanalyse uitgevoerd vanuit het gezondheidszorgperspectief en het maatschappelijk perspectief, waarbij het meest effectieve DMP en het minst effectieve DMP binnen iedere ziektecategorie werden vergeleken. De resultaten vertoonden een grote variatie in kosten voor ontwikkeling en implementatie tussen DMP's (van € 16 tot € 1.709) en onderstreepten het belang van schaalvoordelen. Veranderingen in gezondheidszorgkosten waren niet statistisch significant. Bij alle ziekten werden DMP's in verband gebracht met verbeteringen ten aanzien van de integratie van CVR-zorg (0,10 PACIC-eenheden), lichaamsbeweging (+0,34 weekdagen waarop patiënten ten minste 30 minuten matige of zware lichaamsbeweging hadden) en het stoppen met roken (8% minder rokers). Aangezien een toename van lichaamsbeweging en 'self-efficacy' voorspellend waren voor een verbetering van de kwaliteit van leven, hebben DMP's die gericht zijn op verbetering hiervan een grotere kans effectief te zijn. Wanneer, binnen een ziektecategorie, het meest effectieve DMP vergeleken wordt met het minst effectieve DMP in termen van de EQ-5D, wijst het merendeel van de bootstrapreplicaties (73% tot 97%) op kostenbesparingen. Dit geldt niet voor COPD (21%). De QALY-winst was laag (0,003 tot +0,013) en omgeven met grote onzekerheid. De conclusie van dit hoofdstuk is dat er na één jaar aanwijzingen waren dat er sprake was van verbeteringen in het niveau van integrale zorg voor CVR-patiënten en van leefstijl voor alle ziekten, maar dat voor geen van de ziekten statistisch significante kostenbesparingen werden gevonden. Het heeft waarschijnlijk meer tijd nodig vooraleer de verbeteringen in zorg tot minder complicaties en ziekenhuisopnamen en dus tot kostenbesparingen zullen leiden.

In **hoofdstuk 8** worden lichaamsbeweging, rookstatus, voor kwaliteit gecorrigeerde levensjaren (QALY's) en de jaarlijkse kosten per patiënt van het meest uitgebreide DMP vergeleken met die van het minst uitgebreide DMP in vier ziektecategorieën: primaire CVR-preventie, secundaire CVR-preventie, beide typen CVR-preventie en COPD (n:1.034). Propensity score matching werd gebruikt om de vergelijkbaarheid tussen DMP's te ver-

groten. Er is een twee-jaars kostenutiliteitsanalyse (CUA) uitgevoerd vanuit het gezondheidszorgperspectief en het maatschappelijk perspectief. Er is een sensitiviteitsanalyse uitgevoerd om het effect van de kosten voor de ontwikkeling en implementatie van DMP's op de kosteneffectiviteit in te schatten. De resultaten wezen uit dat bij patiënten in de meest uitgebreide DMP's een grotere toename van lichaamsbeweging optrad (behalve bij de primaire CVR-preventie) en dat in deze patiëntengroep meer personen stopten met roken. De incrementele QALY's varieerden van -0,032 tot 0,038. Vanuit een maatschappelijk perspectief leidde het meest uitgebreide DMP tot een daling van de kosten van primaire CVR-preventie (zekerheid: 57%), secundaire CVR-preventie (zekerheid: 88%) en beide typen CVR-preventie (zekerheid: 98%). Bovendien leidde de implementatie van uitgebreide DMP's tot QALY-winst bij secundaire CVR-preventie (zekerheid: 92%) en COPD (zekerheid: 69%). De conclusie van dit hoofdstuk is dat de meest uitgebreide DMP's voor CVR en COPD in potentie kostenbesparend, effectief of kosteneffectief zijn ten opzichte van de minst uitgebreide DMP's. De uitdaging voor Nederlandse belanghebbenden is de optimale combinatie van interventies te vinden die voor iedere doelgroep het meest geschikt is.

In **hoofdstuk 9** wordt een methodologisch raamwerk ontwikkeld om de toepassing van een multi-criteria besluitvormingsanalyse (MCDA) voor een uitgebreide economische evaluatie van DMP's te vereenvoudigen. Hiervoor zijn eerder ontwikkelde kaders voor de evaluatie van DMP's en verschillende MCDA-methoden onderzocht en is gebruik gemaakt van onze praktijkervaringen met economische evaluatie van DMP's en persoonlijke gesprekken met belanghebbenden in de chronische zorg. Het kader bevat verschillende doelstellingen en criteria die relevant zijn voor de evaluatie van DMP's en indicatoren om te meten hoe goed de DMP's score op deze criteria. In het kader wordt onderscheid gemaakt tussen de ontwikkelingsfase en de implementatiesfase van DMP's. De doelstellingen van DMP's worden ingedeeld in a) veranderingen in het proces van zorgverlening, b) veranderingen in de leefstijl en het zelfmanagement gedrag van patiënten, c) veranderingen in biomedische, fysiologische en klinische gezondheidsuitkomsten, d) veranderingen in gezondheidsgerelateerde kwaliteit van leven, en e) veranderingen in finale gezondheidsuitkomsten. Tevens zijn alle relevante kosten van DMP's in het kader opgenomen. Op basis van dit raamwerk is een MCDA uitgevoerd van een hypothetisch DMP ten opzichte van de gebruikelijke zorg. Dit hoofdstuk roept op tot een bredere economische evaluatie van DMP's die niet op slechts één enkel criterium gebaseerd is maar waarbij rekening gehouden wordt met meerdere relevante criteria tegelijkertijd. Het in dit hoofdstuk gepresenteerde kader is een stap in de richting van standaardisering van een dergelijke evaluatie.

Hoofdstuk 10 bevat een algemene discussie op basis van de voornaamste bevindingen van de voorgaande hoofdstukken. In dit hoofdstuk wordt een gemengde bekostigingssystematiek voorgesteld met een passende combinatie van financiële prikkels

voor zorgaanbieders en zorgbetalers. Het ontwerp van die systematiek moet worden afgestemd op de structuur van het zorgstelsel, en de kosten voor de ontwikkeling en implementatie van DMP's dienen in de bekostiging te worden opgenomen. Verder dient de PFP-component goed functionerende DMP's te belonen en kan een systeem van gezamenlijk gedeelde besparingen "speculatief gedrag" voorkomen en de prikkels voor de belanghebbenden op elkaar afstemmen. Bovendien wordt in dit hoofdstuk gesteld dat het vinden van de juiste combinatie en intensiteit van DMP-interventies die optimaal zijn afgestemd op een specifieke doelpopulatie en met succes worden geimplementeerd, het recept is voor een maximale effectiviteit van DMP's. De implicaties van de bevindingen van het proefschrift worden ook besproken in hoofdstuk 10. Deze hebben vooral betrekking op de juiste definitie en het juiste gebruik van prestatie-indicatoren, het ontwerp van een goed functionerend geïntegreerd ICT-systeem, afstemming van de DMP-interventies op de doelpopulatie en het opnemen van meerdere typen gezondheidszorg in de gebundelde betaling. Tot slot worden in hoofdstuk 10 de methodologische uitdagingen en mogelijke oplossingen alsook suggesties voor toekomstig onderzoek besproken.

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11.4 PhD portfolio

11.4.1 Publications in this thesis

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11.4.3 Oral Presentations in scientific conferences

Costs and effects of new professional roles: evidence from a literature review, International Health Economics Association (iHEA), Dublin (IRL). July 2014

Evidence-based policy making: how cost-effective are disease management programs in the Netherlands? International Conference in Integrated Care, Brussels (B). April 2014

System-wide impact of payment schemes of chronic care: evidence from an empirical analysis , LoLa HESG, May 2013

System-wide impact of payment schemes of chronic care: evidence from an empirical analysis, International Conference in Integrated Care, Berlin (DE). March 2013

Hierarchical modelling of patient's costs in disease management programs for cardiovascular risk, diabetes, and COPD, European Conference on Health Economics (ECHE). Zurich (CH). July 2012.

The use of multi-criteria decision analysis to elicit colorectal cancer screening preferences (Hummel et al., 2012) discussant at 'Low Lands Health Economists' Study Group (LoLa HESG)'. Zutphen (NL). May 2012.

Developing a framework to facilitate the full economic evaluation of disease management programs, presented at 'International Conference in Health Economics, Policy and Management'. Athens (GR). July 2011.

Developing a framework to facilitate the full economic evaluation of disease management programs, presented at 'Low Lands Health Economists' Study Group (LoLa HESG)'. Utrecht (NL). May 2011.

Towards integrated care for chronic conditions: Dutch policy developments to overcome the (financial) barriers, presented at '11th Conference of Integrated Care'. Odense (DEN). March 2011.

11.4.4 Poster Presentations in scientific conferences

Cost-effectiveness of disease management programs for cardiovascular risk and COPD in the Netherlands, International Society for Pharmaceutical Outcomes and Research (ISPOR) 2014, Amsterdam (NL). November 2014

System-wide impact of payment schemes of chronic care: evidence from an empirical analysis, International Society for Pharmaceutical Outcomes and Research (ISPOR) 2012, Berlin (DE). November 2012

Towards integrated care for chronic conditions: Can the new Dutch payment scheme overcome the barriers?, presented at 'European Conference on Health Economics (ECHE) 2010'. Helsinki (FIN). July 2010.

11.4.5 Short courses

Pharmaceutical Pricing- ISPOR, November 2012
Instrumental variables- ISPOR, November 2012
Propensity score matching- ISPOR, November 2012
Repeated measurements- NIHES. March 2011
Problem Based Learning teaching skills- EUR. Januari 2011
Academic writing Academic Language Center- Leiden University. June 2010
Basic course in didactics- EUR. September 2010
Missing values in clinical research- NIHES. April 2010
Klaar in vier jaar (ready in four years) Brigitte Hertz (NL). April 2010

11.4.6 Teaching

Co-coordinator, lecturer and tutor: Pharmaceutical pricing, master programme Health Economics, Policy and Law, Erasmus University Rotterdam. 2012 - 2014.

Tutor: Advanced research methods: master programme Health Economics, Policy and Law, Erasmus University Rotterdam. 2013 - 2014.

Tutor and supervisor: Quantitative Analysis in Health Care, 2nd year BMG Bachelor (in Dutch), Erasmus University Rotterdam. 2010 - 2014.

Lab instructor: Health Economics, short course, Netherlands Institute for Health Sciences (NIHES). 2010 - 2014.

Lab instructor: Health Technology Assessment, Master in Health Economics, Policy and Law, Erasmus University Rotterdam. 2012 - 2014.

Supervisor and co-evaluator: Master thesis, master programme Health Economics, Policy and Law, Erasmus University Rotterdam. 2010 - 2014.

11.5 Curriculum Vitae

Apostolos (1981) was born and raised in Larisa, Greece. He obtained (2004) his bachelor degree in Health Care Management from the Technological Educational Institute of Kalamata after having conducted his graduate research at the Academic Hospital Maastricht. He also obtained a masters degree (MSc) in Health Economics, Policy and Management from the University of Maastricht (2006) and a masters degree (MSc) in Financial Economics from the Erasmus University Rotterdam (2007). After finishing his studies, he worked for two years as consultant at APE b.v., a public economics consultancy located in The Hague. His work at APE b.v. was related to health and labour economics and was commissioned by the Dutch government, the European Commission, and private companies. In June 2009, Apostolos joined the Institute for Medical Technology Assessment (iMTA), department of Health Policy and Management (iBMG), Erasmus University Rotterdam where he got involved in several research projects related to the economic evaluation of integrated care. After having completed his PhD research, he continued his career as senior researcher at the Health Economics Research Centre (HERC), University of Oxford.

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