Health-economic evaluations of person-centred care

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Abstract

Person-centred care aims at making the individual partake in the healthcare decision-making and at supporting individual health management. This stands in contrast to usual care, which typically has more focus on the particular disease at hand, rather than on the person behind the disease. Interventions in which care is delivered according to the person-centred care approach belong to a larger group of interventions, usually referred to as complex interventions. It is well-known that evaluating such interventions frequently entails methodological challenges. The overall objective of this thesis was to contribute to the field of evaluation of complex interventions, by adding to the emerging, but still rather scarce, knowledge concerning the effects and the cost-effectiveness of person-centred care interventions. An essential part of this endeavor was to examine the effects achieved by person-centred care by applying a range of different outcome measures and methods.

The thesis is comprised of four articles, all of which employed data from randomized controlled trials of person-centred care interventions conducted at the University of Gothenburg Centre for Person-Centred Care. In study I the effects of a person-centred care intervention for patients with acute coronary syndrome was estimated. In studies II and III, the cost-effectiveness of person-centred care provided to patients with (i) acute coronary syndrome and (ii) chronic obstructive pulmonary disease and/or chronic heart failure, compared with usual care, was estimated. In study IV, the outcomes observed among patients with acute coronary syndrome receiving person-centred care, or usual care, were projected to a post-trial point in time. A Markov-type health-economic model was constructed and the corresponding long-term cost-effectiveness of person-centred care was calculated. Overall, the results obtained in these studies suggest that person-centred care is both more effective and less costly than usual care, both in the short and in the long-term perspective.

Keywords: Person-centred care; Economic evaluation; Cost-effectiveness; Health outcomes; Markov model; Randomized controlled trial; Acute coronary syndrome; Chronic heart failure; Chronic obstructive pulmonary disease

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Sammanfattning på svenska

Begreppet person-centrerad vård har använts en längre tid och vårdfilosofin har implementerats på ett flertal sjukhus. Person-centrerad vård innebär att patienten är delaktig i den egna vården och att en omsorgsetik tillämpas där patienten bemöts som en person, och inte sin sjukdom. Den vetenskapliga litteraturen rapporterar ett relativt fåtal forskningsresultat avseende effekterna och kostnadseffektiviteten av person-centrerad vård. Det övergripande syftet med de studier som utgör föreliggande avhandling var att bidra till denna forskning genom att undersöka effekterna av och kostnadseffektiviteten hos person-centrerad vård, jämfört med traditionell vård, för specifika sjukdomskontexter. I någon mån bidrar studierna även till att förbättra befintliga hälsoekonomiska utvärderingsmetoder genom att belysa hur dessa lämpligast tillämpas på området person-centrerad vård.

Studierna använder primärdata som insamlats inom ramen för två randomiserande kontrollerade studier vid Göteborgs Universitets Centrum för person-centrerad vård. Dessa kliniska studier gäller personer med akut koronart syndrom respektive personer med kronisk obstruktiv lungsjukdom och/eller hjärtsvikt. I den första delstudien undersöktes effekterna av person-centrerad vård, jämfört med traditionell vård, hos personer med akut koronart syndrom. Analyser genomfördes på hälsorelaterade utfall i ett antal dimensioner: hälsorelaterad livskvalitet, tilltro till sin egen förmåga, återgång till arbete och fysisk aktivitet. Patienterna i interventionsgruppen visade högre tilltro till sin egen förmåga än patienterna i kontrollgruppen. Samma patienter hade även högre hälsorelaterad livskvalitet, återgick till arbetet och till tidigare nivå av fysisk aktivitet i större grad jämfört med gruppen som inte fick person-centrerad vård.

I avhandlingens andra delstudie jämfördes kostnader och effekter mellan person-centrerad vård och traditionell vård för personer med akut koronart syndrom. De genomförda kostnadseffektivitetsanalyserna visade betydande skillnader för personer yngre respektive äldre än 65 år. Analyserna tyder på att person-centrerad vård var både kostnadsbesparande och mer effektiv än traditionell vård för personer yngre än 65 år. Däremot var den traditionella

vården både kostnadsbesparande och mer effektiv än person-centrerad vård för personer 65 år och äldre.

I den tredje delstudien analyserades kostnadseffektiviteten hos person-centrerad vård, jämfört med traditionell vård, för personer med hjärtsvikt och/eller kroniskt obstruktiv lungsjukdom. Kostnadseffektivitetsanalyserna visade att person-centrerad vård var både kostnadsbesparande samt mer effektiv jämfört med traditionell vård, för hela studiepopulationen men även för studiepopulationen utan de patienter som avled under studieperioden.

Den fjärde delstudiens huvudsyfte var att undersöka den långsiktiga kostnadseffektiviteten hos person-centrerad vård, jämfört med traditionell vård, för patienter med akut koronart syndrom. I detta syfte utvecklades en hälsoekonomisk simuleringsmodell som replikerar den kliniska situationen vid akut koronart syndrom. Med hjälp av denna modell skrevs den tillgängliga kliniska datan fram från ett tvåårsperspektiv till ett femårsperspektiv. Modellen utnyttjar en kombination av primärdata och information som inhämtats dels från nationella regionala register och dels från den vetenskapliga litteraturen. Kostnadseffektiviteten hos person-centrerad vård beräknades både för ett tvåårs- och för ett femårsperspektiv. Resultaten tyder på att de positiva effekter som observerades för det tvååriga tidsperspektivet även sträcker sig till ett femårsperspektiv.

Sammanfattningsvis tyder de resultat som uppnåtts inom ramen för de fyra ovan sammanfattade studierna på (1) att person-centrerad vård har betydande effekter på hälsorelaterad livskvalitet (och ett antal andra mått på hälsotillstånd), (2) att person-centrerad vård potentiellt är kostnadsbesparande genom effekter på det medicinska vårdutnyttjandet och sjukskrivningar, (3) att person-centrerad vård är att föredra framför traditionell vård ur ett kostnadseffektivitetsperspektiv, åtminstone för de som behandlas för akut koronart syndrom och kroniskt obstruktiv lungsjukdom och/eller hjärtsvikt, och (4) att kostnadseffektiviteten hos person-centrerad vård kvarstår då tidsperspektivet förlängs bortom existerande kliniska studier.

List of papers

I

Pirhonen L., Olofsson E.H., Fors A., Ekman I., Bolin K.

Effects of person-centred care on health outcomes-A randomized controlled trial in patients with acute coronary syndrome

Health Policy, 2017. 121(2): 169-179.

II

Pirhonen L., Bolin K., Olofsson E.H., Fors A., Ekman I., Swedberg K., Gyllensten H.

Person-centred care in patients with acute coronary syndrome: cost-effectiveness analysis alongside a randomized controlled trial

PharmacoEconomics – Open, 2019. 3(4): 495-504.

III

Pirhonen L., Gyllensten H., Olofsson E.H., Fors A., Ali L., Ekman I., Bolin K.

The cost-effectiveness of person-centred care provided to patients with chronic heart failure and/or chronic obstructive pulmonary disease

Submitted

IV

Pirhonen L., Gyllensten H., Fors A., Bolin K.

A health-economic model of competing interventions for patients with acute coronary syndrome

Submitted

Content

Abbreviations

ACS • Acute coronary syndrome
ADL • Activity of daily living
BMI • Body mass index
CBA • Cost-benefit analysis
CEA • Cost-effectiveness analysis
CEAC • Cost-effectiveness acceptability curve
CE • Cost-effectiveness (plane)
CHF • Chronic heart failure
COPD • Chronic obstructive pulmonary disease
CUA • Cost-utility analysis
DRG • Diagnosis related groups
GPCC • University of Gothenburg Centre for Person-centred Care
GSE • General self-efficacy
ICECAP • Investigating Choice Experiments Capability Measure
ICER • Incremental cost-effectiveness ratio
MI • Myocardial infarction
MiDAS • Mikrodata för Analys av Socialförsäkringen
NE • North-east (quadrant)
NW • North-west (quadrant)
OLS • Ordinary least squares
PMM • Predictive Mean Matching
QALY • Quality-adjusted life year
RCT • Randomized controlled trial
SE • South-east (quadrant)
SPDR • Swedish Prescribed Drug Register
SW • South-west (quadrant)

Introduction

The concept of person-centred care has been in use for some time [1] both among healthcare practitioners and, to some extent, in the research community. However, it has not received much attention from health-economic researchers. Efficient use of scarce resources necessitates that costs and effects are compared between alternative and competing usages. More specifically, costs and the cost-effectiveness of person-centred care practices are essential pieces of information when considering the implementation of such practices in the healthcare system [2]. Also, this information is necessary when evaluating new care models, such as person-centred care, due to the mandatory cost-effectiveness principle [3]. Thus, health-economics is an essential aspect in healthcare science. Typically, results from health-economic evaluations are less than clear cut, even in cases where a well-defined intervention, for instance a new pharmaceutical treatment, is studied and effectiveness data from randomized controlled trials are available. This is due to uncertainty concerning the costs and effects induced both by the intervention at hand and by its comparison. In several real-life situations, however, the intervention may not be as well-defined as in the pharmaceutical comparison case. Person-centred care interventions belong to a larger family of interventions usually referred to as complex interventions, with the common feature of not being as standardized regarding design and/or delivery as, for instance, a drug-based intervention [4]. For this reason, it is not too surprising that there seems to be too little available information on whether or not person-centred care is cost-effective compared to usual care in different specific settings and for different specific diseases [5-7].

Treatment effects reported in studies of person-centred care differ not only since different diseases are studied, but also vary according to study end points and the empirical measurements of those endpoints. Moreover, the current state of knowledge concerning the effectiveness of and the interaction among the individual components constituting the total person-centred care intervention is inadequate. In addition, the assessment of the effects of person-centred care interventions from published studies is aggravated by the fact that (1) there seems to be no consensus in the scientific literature as to the exact definition of person-centred care and, hence, published results pertain to a variety of different interventions more or less adhering to the core features of interventions from the

University of Gothenburg Centre for Person-centred Care (GPCC), (2) there is a large variability in the outcome measures used and, (3) frequently, it is not strictly structured person-centred interventions (such as GPCC) that have been evaluated [7-10]. In many cases, the studied intervention contains only parts of the building blocks of a GPCC intervention, and is labelled as, for instance, self-management programs or patient-centred care.

The overall objective of this thesis was to improve available knowledge concerning the effects and the cost-effectiveness of person-centred care interventions. Needless to say, only a small fraction of the therapeutic areas in which the current knowledge prevents, or at least aggravates, the implementation of cost-effective person-centred care approaches is covered.

Background

In this section, the current knowledge regarding the effectiveness and the cost-effectiveness of person-centred care and of other healthcare interventions with the patient in focus will be summarized. The section starts by defining patient-centred and person-centred care and, complex interventions, a broader classification into which the different types of interventions included can be classified. Apart from interventions based on patient- and person-centred care, interventions labelled as disease management and self-management programs were included, since these interventions have some components in common with the former class of interventions. The search for literature was not intended to be performed as a complete systematic review (see the appendix for details).

Complex interventions

A complex intervention is defined as an intervention comprised of several interacting components, for example, a number of different behaviors required by the ones delivering and/or receiving the intervention, several groups targeted by the intervention, flexibility in the intervention delivered and multi-dimensional outcomes [4, 11]. Person-centred care interventions always include one, or several, of these components. Person-centred care interventions are often delivered on several healthcare levels, they require different behaviors by the ones delivering and receiving the intervention and flexibility is required when delivering the intervention.

Person-centred care

Healthcare is increasingly being delivered using a patient-centred approach rather than a disease-focused approach [12-14], and in recent years the implementation of person-centred care has become more frequent [12]. There are considerable differences between the attempts that have been made in order to provide healthcare tailored for the individual patient. Hence, only a subset of those attempts may resemble, or coincide, with the specific definition of person-centred care. More specifically, patient-centred care aims at acknowledging the

patient as an individual and not viewing the patient through a biopsychosocial perspective. This is different from person-centred care, which originates from a philosophy of care emanating from the needs of the individual, and aims at accentuating the patient as a unique person with individual needs, resources and goals. Moreover, person-centred care strives at creating a partnership between the patient and healthcare professionals. Thus, patient-centred care and person-centred care are not identical concepts [12-16]. Studies on patient-centred care and other interventions that focus on the patient potentially have implications that are transferable to a person-centred care setting. Thus, the empirical results reported in the literature for patient-centred interventions are potentially significant also for interventions based on person-centred care.

The interventions evaluated in this thesis originate from the University of Gothenburg Centre for Person-centred Care (GPCC). According to the GPCC approach, there are three well-defined components that need to be present in order for provided healthcare to be classified as person-centred care: 1) the patient tells his or her patient narrative (a description of the patient's own perception of his or her illness, symptoms, the influence these have on life, goals after hospitalization discharge and resources available to reach these goals), 2) developing a partnership, leading to an agreement between the patient and his or her caregivers about how to reach the goals set up by the patient and based on these goals constructing a health plan, and 3) documenting, agreeing on, and signing the patient narrative and the health plan. Writing down the narrative and health plan gives legitimacy to the patient's story. The health plan can at any point in time be updated and/or followed-up by either the patient or the caregivers, or both, for example, during a re-hospitalization [12]. The three steps in the GPCC approach are illustrated in Figure 1, below.

Figure 1. Steps in the GPCC approach.

Reported quality of life and functional effects of person-centred and patient-centred care

Person-centred and patient-centred care have been reported to influence various healthcare outcome measures, for instance, quality of life, self-efficacy, and physical and psychological functioning positively, which has been demonstrated in several studies and for several therapeutic areas [7, 10, 17, 18]. For instance, such effects of person- or patient-centred care have been found, when provided to patients with various chronic diseases: (1) hearth failures/heart diseases [6, 19-26], (2) diabetes [27-29], and (3) chronic conditions in general [30-34]. In what follows, a more detailed description, by health condition, of published studies on the effects of person- and patient-centred care will be provided. An overview is provided in table A1 (the appendix).

Heart failure and heart diseases

The effects of person-centred care provided to patients with heart failure have been examined in a number of studies and it has been demonstrated to improve several outcomes. A 2015 systematic review concluded that person-centred care provided to patients with heart failure improves quality of life, physical and mental status, and self-efficacy [21]. In another 2015 review, it was concluded that patient-centred care provided to patients with chronic heart failure improves health-related quality of life, reduces symptom burden and mitigates depression [24]. Moreover, it has been found that disease management programs reduce healthcare costs, improve disease coping, and increase health-related quality of life [19, 35]; that person-centred care reduces healthcare costs compared to conventional care (a reduction of hospital stays by 30 percent has been reported), improves activity of daily living (ADL) functioning, improves quality of life and reduces re-hospitalizations [6, 22, 25, 36]; that self-care interventions improve medication adherence [20]; and that specialist nurse management programs reduce the number of hospital readmissions [37]. The evidence in support of a positive effect on health of interventions included here are not completely unambiguous though: a 2015 randomized controlled trial of a patient-centred disease management program provided to patients with heart failure, did not find any improvements in general health [38].

Diabetes

There are numerous studies on the effects of patient-centred care provided to patients with diabetes. The findings include, for example, that a patient-centred

care intervention leads to greater patient satisfaction with the healthcare provided, fewer symptoms of depression, fewer days in bed due to illness and greater self-efficacy [27]; that patient-centred care reduces HbA1c up to two years after the intervention [28]; and that strengthening patient involvement improves diabetes outcomes (for example diabetes control and knowledge of the disease) and patient self-care [29]. Other studies, however, suggest that patient-centred care and disease management programs have no effect on health-related quality of life among diabetes patients [39-42].

Chronic conditions in general

In this section, a summary is given on studies that investigate the effects of interventions for patients with chronic conditions in general (without necessarily differentiating between the specific conditions). Health interventions delivered according to a patient-centred care approach, and provided to patients with one or more chronic condition, for instance, hypertension or diabetes, have been found to produce positive effects beyond those attained using a standard-care approach. More precisely, it has been demonstrated that a patient-centred based delivery of healthcare improves self-efficacy, and that the improvement is most pronounced among those with the largest number of health conditions and/or with the most severe diseases [30]. Further, collaborative care efforts provided by different care givers aimed at patients with depression and diabetes and/or coronary heart disease, have been found to improve medical outcomes, reduce depression, and increase quality of life [31]. Disease management and self-management programs have also been demonstrated to improve health, to improve health-promoting behaviours, and lead to fewer days of hospitalization as well as fewer hospitalizations, compared to standard care [32, 33, 43]. In contrast, however, some evidence suggest that patient-centred care has little or no impact on health-related outcomes [44, 45]. A 2013 systematic review on the effects of different patient-involvement interventions, such as patient education, self-care management and patient involvement, concluded that these types of interventions frequently had little impact on clinical, functional, and emotional outcomes [10]. Another systematic review found patient-centred care to be ineffective when delivered to patients with chronic diseases [46]. Similarly, a systematic review on personalised care planning, including interventions emphasizing shared-decision making, provided to patients with chronic diseases did not identify any effects on health-related quality of life [47].

Other diseases

Patient- and person-centred care, as well as disease management programs and self-management programs, have been studied in a number of other disease or health-status contexts, and have been demonstrated to decrease healthcare costs [48] and potentially increase health-related quality of life [49] for patients with head and neck cancer, to increase quality of life for patients with HIV [50], to improve patient outcomes, such as quality of life and asthma control, in asthma care [51], to improve self-efficacy among patients with chronic kidney disease [52] and among patients with depression or anxiety [53], to reduce the length of hospital stay among hip fracture and total hip arthroplasty patients [54, 55], to improve quality of life, self-efficacy and ADL among stroke patients [56-58], and to improve health-related quality of life among patients with dementia [59-62], among patients with depression [63], among patients who have survived gynaecological cancer [64], and among persons with intellectual disabilities [65]. In contrast, inconclusive effects of patient-centred care on outcomes have been reported among patients in primary care [5], and on health-related outcomes among children with chronic diseases [66].

Cost-effectiveness of person-centred care

The number of peer-reviewed publications on the cost-effectiveness of complex interventions in general, and on person-centred care in particular, is relatively small. The studies that have been published are mostly confined to chronic diseases, such as chronic heart failure and acute coronary syndrome. Person-centred care has been found to be cost-effective for patients with hip fracture [67], heart failure [6], head and neck cancer [48], and acute coronary syndrome [68]. The cost-effectiveness evidence regarding disease management and patient-centred care is more comprehensive. The evidence is mixed, however, as some studies find these programs to be cost-effective [69-72] while other studies suggest the opposite [73].

While the time range of clinical studies in most cases is between 1 and 3 years, the effects of the studied interventions typically stretch over a longer time period. Thus, in order to examine the cost-effectiveness of health interventions entailing effects that stretches beyond the end of corresponding clinical studies, the costs and the effects induced by the particular intervention must be projected beyond this point in time. Typically, this is achieved by employing some type of mathematical modelling, using data from the clinical study in combination with additional sources of information. The purpose of health-economic modelling is

to integrate the information from different sources into a unified framework used for calculating specific outcomes, for instance, an incremental cost-effectiveness ratio pertaining to a specific utilization of resources as compared to a competing allocation [74, 75].

Few studies have been published on the long-term cost-effectiveness of patient- or person-centred care. The studies that have been published are mainly confined to interventions directed towards patients with diabetes and cardiovascular conditions. For instance, patient-centred care provided to patients with type 2 diabetes has been found to be cost-effective in a lifetime perspective [76, 77]. Cleveringa et al. [78] studied the long-term effects of a diabetes management intervention, aiming at reducing cardiovascular risks, using microsimulation modelling. They found that the intervention was more costly and produced a slight improvement of health compared to usual care and was only cost-effective for a subgroup of patients. Further, a recent study on patients with heart failure found, employing a Markov-modelling approach which projected the clinical study outcomes to a 10-year period, that a multidisciplinary disease management program was cost-effective compared to standard care [79].

Studied health conditions

In this section, the three diseases studied in this thesis are described.

Acute coronary syndrome

Acute coronary syndrome (ACS) is a disease which includes a number of acute myocardial ischemic states, specifically unstable angina, ST segment elevation infarction and non-ST segment elevation infarction [80]. Mortality among patients with ACS is elevated, both during hospitalization and several years after discharge [80, 81]. The five-year all-cause mortality rate after an ACS event has been reported to be as high as 40 % [82]. Further, patients with ACS have a high risk of re-hospitalization, both in the short and long-term. It has been reported that 30 % of ACS-patients are re-hospitalized within 6 months after their first ACS event and that one in five patients will experience an ACS-related event within the first 5 years after the initial ACS-event [83, 84]. The high frequency of re-hospitalizations leads to considerable direct costs both during and immediately after hospitalization, but also as long as 5 years after discharge [85-87]. In addition to large direct costs, ACS is also associated with considerable indirect costs due to sickness absenteeism. Productivity losses are a substantial part of

the economic burden of the disease and because of these losses ACS has been reported to cause larger costs to employers than other common diseases [86, 88].

Chronic obstructive pulmonary disease

Chronic obstructive pulmonary disease (COPD) is a slowly progressive condition and comprises of a range of chronic respiratory diseases which are characterized by airflow limitation [89, 90]. COPD has a strong association to cigarette smoking [89]. Patients with COPD suffer from a reduction in health-related quality of life [91] together with a 3.5 times higher all-cause mortality rate compared to the general population [92]. COPD generates considerable direct and indirect costs and is considered to be a large problem worldwide [93]. The indirect costs are substantial and have been reported to constitute the main part of the economic burden of COPD for patients still in the workforce [94, 95]. The disease is also characterized by several co-morbidities, for example, cardiovascular diseases, which increases the economic burden further [96-99].

Chronic heart failure

Chronic heart failure (CHF) is a result of abnormalities in the cardiac structure and/or functioning and the symptoms include, for example, fatigue, breathlessness and ankle swelling [100]. CHF is an increasing global health problem and the disease is associated with long hospital stays and high readmission rates [101, 102]. Patients with CHF have been reported to face an in-hospital mortality rate as high as 30 % [103], and a one-year mortality rate at 60 %, for patients with severe CHF [104]. Hospitalizations have been estimated to account for two-thirds of the total healthcare expenditures associated with CHF [104]. The economic burden of CHF is characterized by significant direct and indirect costs [105-107].

Theoretical and methodological framework

Demand for health

Individual health outcomes are determined both by exogenous individual characteristics and by individual choices. A comprehensive empirical health-economic literature demonstrates that the resulting individual health outcomes are partly determined by demographic and socio-economic characteristics [108]. Thus, the choice of demographic and socio-economic explanatory variables employed in study I was based on the health-economic demand-for-health framework developed by Grossman in the early 1970s [109]. The model asserts that the individual to some extent is able to influence his or her own health. More formally, the individual is assumed to "produce" increments to his or her health using an individual capability (a production function) of transforming time and resources into health investments. The individual capability determines how effectively available resources can be transformed into health improvements, i.e., the effective cost of health investments varies between individuals. The individual effectiveness in the "production" of increments to his or her health depends on various individual traits, such as genetics, education and the socioeconomic context. Further, different behaviours "produce" positive or negative increments to health, for instance, wholesome diets and adequate physical exercise produce positive increments to health, while smoking produces negative increments.

Health-economic evaluation

The overall objective of a health-economic evaluation is to compare costs and consequences of competing allocations of available resources [110]. There are two main approaches for performing a health-economic evaluation: the cost-benefit analysis (CBA) and the cost-effectiveness analysis (CEA). Cost-benefit analysis has a solid theoretical foundation in economic welfare theory, while cost-effectiveness analysis has not. The cost-effectiveness approach has been adopted in the majority of published health-economic evaluations, due to very

demanding information requirements associated with the cost-benefit approach [111]. Essentially, the cost-benefit approach requires knowledge about the total individual willingness to pay for the studied competing allocations of resources, i.e., including costs and benefits pertaining not only to health outcomes [112].

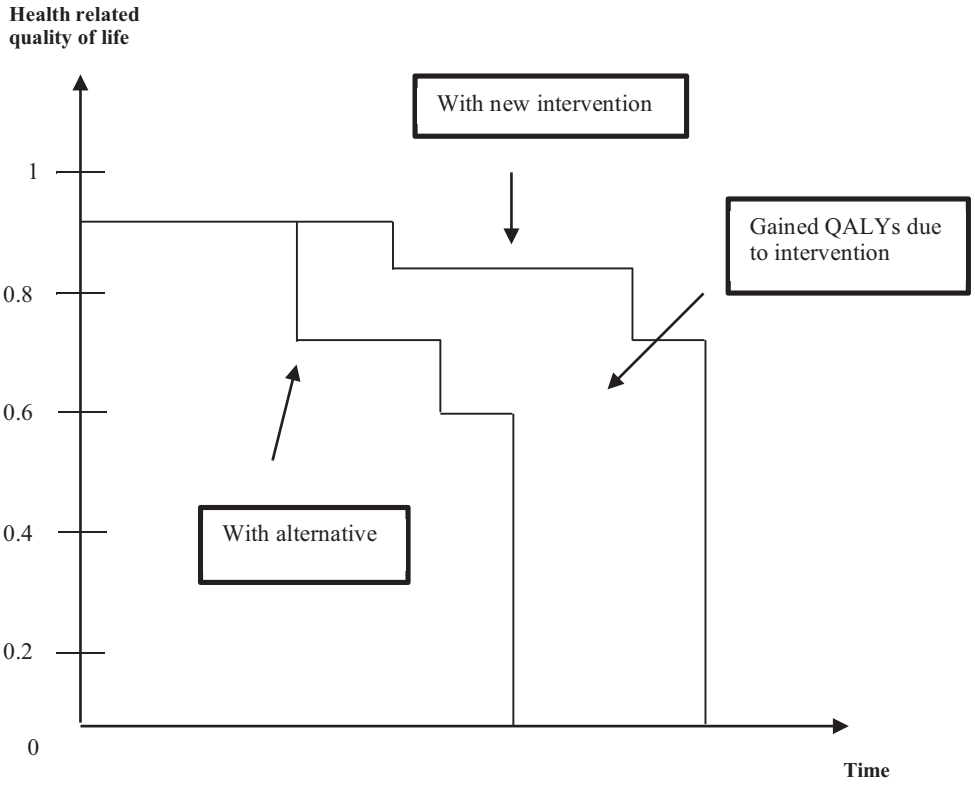
Cost-effectiveness analysis

Cost-effectiveness analysis entails less demanding information requirements, compared to cost-benefit analysis, as it compares observable, or relatively readily measurable, outcomes to the costs associated with the efforts employed to reach those outcomes. Cost-effectiveness analyses employing utility measures of attained health-related outcomes are often labelled as cost-utility analyses (CUA). The most employed cost-effectiveness measure is the incremental cost-effectiveness ratio (ICER), which is the marginal cost of achieving a one-unit increase of the effect. More formally, it is defined as follows:

$$ICER = \frac{Cost_a - Cost_b}{Effect_a - Effect_b} = \frac{\Delta Cost}{\Delta Effect}$$

The dominating health outcome measure employed in cost-effectiveness analyses is the quality-adjusted life year (QALY). A QALY is a composite measure of quality of life and length of life, and, hence a difference in QALYs attained by two competing treatment alternatives reflects a combination of differences in quality of life and in expected length of life (or survival within the time horizon applied) [110]. Figure 2 illustrates how a QALY is calculated.

Figure 2. The calculation of a QALY.



QALY = Quality-adjusted life year. The area under each particular graph is the number of QALYs associated with that particular resource allocation.

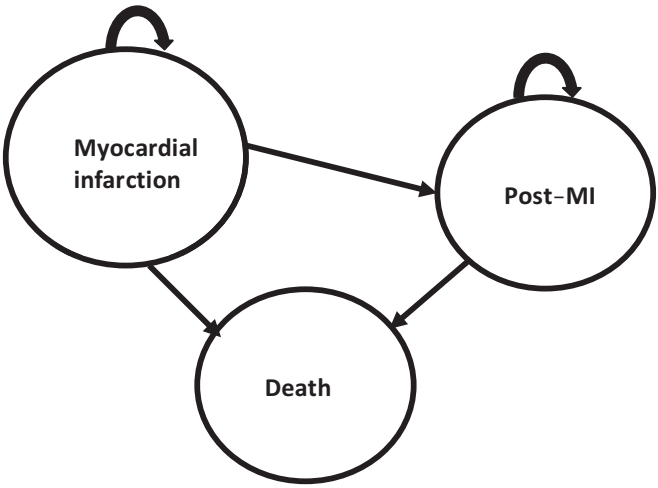
Health-economic modelling

Frequently, health-economic evaluations apply time perspectives that go beyond the reach of available clinical trial data. In such cases, the clinical trial data regarding costs and outcomes need to be projected to the appropriate timeframe. In such cases, mathematical modelling is used for projecting data which is not (yet) empirically available. The most frequently employed type of mathematical model in health-economic evaluation belongs to a class of models referred to as Markov models. They exhibit the convenient Markov property, which means that the future state of a simulated system depends only on the current state of the sys-

tem. This simplifies the necessary calculations considerably, from being, in most case, relatively intractable to being handled rather straightforwardly.

A health-economic Markov model consists of a finite set of health states where the patient moves between these states based on different transition probabilities. It is important to note, that although the progress of a modelled disease is stochastic, the actual Markov-based calculations are based on given probabilities and, hence, are deterministic. Transition between health states occurs at fixed points in time. The length of time between two instances in which transitions between states occur is called a Markov Cycle. The cycles need to be short enough to capture important events in a disease progress but long enough to simplify the structure of the model. Moreover, the health states need to reflect significant clinical and economical events associated with the specific health condition(-s) studied. Using available estimates of, for instance, resource utilization and quality of life associated with each health state, the Markov model produces projections of total costs associated with competing resource allocations, and for the modelled time horizon [75, 112]. The uncertainty of the projected costs and effects can be inferred either by Monte Carlo simulation or by a bootstrapping approach. Monte Carlo simulation amounts to specifying stochastic processes according to which the variables included in the model are generated, and to execute the Markov model repeatedly. Bootstrapping requires access to primary data and is basically executed by repeated executions of the Markov model, using different subsets of the clinical data. In figure 3 a simple Markov structure is illustrated. In the model, an individual (or all individuals in a population) has a myocardial infarction (MI) in the first cycle, and may, after the first cycle, make a transition to one of three mutually exclusive states: a new infarction, a post-MI state or death. When the individual arrives at the post-MI state he or she stays there until death. Death is, obviously, an absorbing state, which means that once an individual is in that state, he or she cannot leave that state.

Figure 3. Example of a simple Markov structure



MI = Myocardial infarction

Aims

The overall aims of this thesis were (a) to improve and extend available and applicable knowledge of the cost-effectiveness of particular person-centred care interventions and (b) to improve and extend the methods for performing health-economic evaluations in the area of person-centred care, by exploring the usefulness of health-economic methods and various outcome measures in different disease and institutional contexts.

The thesis is comprised of four studies utilizing primary data from two randomized controlled trials (RCT) involving patients with (1) ACS, and (2) COPD and/or CHF. The specific objectives in each paper were the following:

Study I: To analyse the effects of person-centred care, directed towards patients with ACS, on four different measures of health-related outcomes: (1) health-related quality of life, (2) general self-efficacy, (3) physical activity, and (4) return to work.

Study II: To analyse the cost-effectiveness of person-centred care, directed towards patient with ACS, as compared to usual care. The analysis was performed for the time horizon of 1 year.

Study III: To analyse the cost-effectiveness of person-centred care, directed towards patients with COPD and/or CHF, as compared to usual care. The time horizon for the cost-effectiveness analysis was 6 months.

Study IV: To estimate the long-term cost-effectiveness of person-centred care, compared to usual care, provided to patients with ACS, by constructing a simulation-model for projecting outcomes beyond the reach of the clinical data.

Data and methods

The clinical trials

Below, a detailed account of the clinical trials from which the data was collected is presented.

The ACS trial (study I, II and IV)

The data employed for study I, II and IV was collected from a RCT of a person-centred care intervention directed towards patients with ACS [113, 114]. Patients were recruited between June 2011 and February 2014 at two sites at the Sahlgrenska University Hospital, Gothenburg, Sweden. The intervention was delivered on three healthcare levels: inpatient care, outpatient care, and primary care. The persons included in the trial and randomized to either person-centred care or usual care were hospitalized due to ACS. The intervention and control group consisted of 94 and 105 patients, respectively. The inclusion criteria were: 1) age below 75 years, and 2) hospitalized for ACS (ICD-10 code I200, I209 or I21). The following exclusion criteria were applied: 1) currently listed at a private primary care centre or at a primary care centre in another region; 2) no permanent address; 3) planned for heart surgery (e.g. coronary artery bypass grafting [CABG]); 4) cognitive impairment; 5) alcohol and/or drug abuse; 6) survival expectancy less than one year or; 7) participating in a conflicting study.

Prior to the trial, the healthcare personnel at the sites where the intervention was implemented were informed, at seminars and workshops, about how to integrate the person-centred way of delivering care into their work. During the trial, booster-seminars were arranged, at which the personnel had the opportunity to discuss and share thoughts about the implementation of the intervention. The control group received usual care. The intervention group also received usual (medical) care and, in addition, the GPCC approach, where the three person-centred care steps described earlier were covered. A health plan was created based on the patient narrative and attainable goals that were agreed upon by both the patient and the healthcare professionals. Primary care was provided at separate units for the control and the intervention group. Those randomized to the intervention group received healthcare by the GPCC team at the primary care

centers when the patients visited a primary care center. Those randomized to the control group received usual care from primary care personnel at other primary care centers.

The data

At baseline, data on demographic and socio-economic characteristics, such as age, gender, education, employment, and income was collected together with information on disease-specific characteristics, for example, previous MI, severity of current infarction and heart-related treatments at hospital. Moreover, the patients were supposed to fill out a series of questionnaires comprising of questions about, for example, health-related quality of life and general self-efficacy. Responses to the questionnaire were collected at baseline, and thereafter at 4 weeks, 8 weeks, 6 months, 1 year and 2 years. The primary endpoint of the intervention was a composite score consisting of self-reported general self-efficacy, return to work or previous activity level and re-hospitalization or death at 6 months after randomization.

The information collected directly from the participants was complemented with register data on pharmaceutical prescriptions, sick-leave, cause of death and healthcare utilization (primary, inpatient and outpatient care) (the Swedish National Board of Health and Welfare and the regional patient register (VEGA; Västra Götaland, Sweden) and the Swedish Social Insurance Agency).

The study population

During the first year after inclusion, 7 patients died; 5 in the intervention group and 2 in the control group. Since expected survival at baseline <1 year was an exclusion criteria in the ACS trial, and since person-centred care is assumed to have no direct effect on mortality, the deceased patients were excluded from study IV and in subgroup analyses in study II.

The COPD/CHF trial (study III)

The data employed in study III was collected from a RCT directed towards patients with COPD and/or CHF [115]. Patients were recruited during 2016 at Sahlgrenska hospital and Östra hospital, Gothenburg, Sweden.

The patients included in this trial and randomized to either person-centred care or to usual care were hospitalized due to relapse and/or worsening of symptoms

due to COPD and/or CHF. The intervention group consisted of 103 patients and the control group of 118 patients. The following inclusion criteria were applied: 1) history of confirmed diagnosis of COPD and/or CHF; 2) admission to hospital for relapse or worsening of symptoms due to COPD and/or CHF; 3) age 50 or above; 4) ownership of a phone with a current subscription to a telephone service provider. Criteria for exclusion were: 1) severe hearing impairment; 2) no registered address; 3) expected survival under a year; 4) cognitive impairment; 5) ongoing documented abuse of alcohol or drugs; 6) other diseases that could interfere with the follow-up; and 7) participation in another randomized study.

The nurses involved in the intervention attended a workshop where they received information about living with COPD/CHF, communication techniques and about how person-centred care should be delivered. The control group received usual care. The intervention group received person-centred care by phone, as an add-on treatment to usual care. After returning home from the hospital the patient received a phone call by the treating nurse on a previously scheduled time. During the phone call, a health plan was created. Thereafter, follow-up phone calls were scheduled. The initial phone call, and follow-up phone calls, served as a forum for discussing specific issues that had arisen, and thoughts in general about the health plan and personal goals.

The data

At baseline, information on demographic and socio-economic characteristics, such as age, sex, and marital status, was collected together with information on disease-specific characteristics, such as diagnosis and previous medical history. In addition, health-related information was collected on, for instance, health-related quality of life, self-efficacy, anxiety and depression, through questionnaires. The questionnaires were distributed at baseline, and thereafter at 3 months and at 6 months.

The information collected from the participating patients was complemented with register data on disposable household income, attained schooling and whether the person was born in Sweden or not (Statistics Sweden), and on healthcare utilization, cause of death and prescribed pharmaceuticals (the Swedish National Board of Health and Welfare and the regional patient register (VEGA; Region Västra Götaland, Sweden)).

The study population

The patients participating in this trial were relatively old (the mean age was 79 at inclusion) and frequently suffered from co-morbidities. During the study period, 37 patients died; 16 in the intervention group and 21 in the control group. In order to avoid significant loss of information, the deceased patients were included in the main analyses in study III. Corresponding calculations were performed excluding the deceased patients, also in study III.

Method specification

In this section, a brief account of the methods used in the four studies is provided. The data and methods used in each paper is summarized in table 1.

Demand for health (study I)

The specification of the regression model estimated in study I, was based on the demand-for-health framework [109]. Four regressions were estimated, employing different empirical measures of health capital: health-related quality of life, self-efficacy, physical activity and return to work. The demand-for health model suggests that health is partly determined by factors such as age, civil status, education, income, and health-related behaviours and individual characteristics, for example, smoking, body mass index (BMI) and the severity of present health conditions. The choice of explanatory variables reflects this.

Short-term cost-effectiveness analysis (study II, study III)

Cost-effectiveness analysis was used in study II and III. Cost-effectiveness measures of person-centred care as compared to usual care were calculated for a number of different alternatives, including different time perspectives and different populations. Moreover, the cost-effectiveness calculations in study II were performed for a societal perspective (complemented with calculations employing a healthcare provider perspective), while the calculations in study III were performed from a healthcare provider perspective only. Costs and effects were not discounted in neither study, due to the short time perspective.

Table 1. Summary of data and methods in papers I-IV

	Study I	Study II	Study III	Study IV
Trial data used	ACS trial	ACS trial	COPD/CHF trial	ACS trial
Register data used	-	Cause of death register SPDR Patient register MiDAS	Cause of death register SPDR Patient register LISA-database	Cause of death register SPDR Patient register MiDAS
Primary data	Socio-economic variables Disease-specific variables Effect data	Socio-economic variables Disease-specific variables Effect data	Socio-economic variables Disease-specific variables Effect data	Socio-economic variables Transition probabilities
Data from registers	-	Cost data Information on deaths	Cost data Information on deaths Socio-economic variables	Cost data
Data from the literature	-	-	-	Effect data Transition probabilities
Methods used	Regression Analysis	Cost-effectiveness analysis	Cost-effectiveness analysis	Markov model
Time frame	6 months 1 year	1 year	6 months	2 years 5 years
Perspective	-	Healthcare provider Societal	Healthcare provider	Healthcare provider Societal

Abbreviations: SPDR=Swedish Prescribed Drug Register; MiDAS=Mikrodata för Analys av Socialförsäkringen; ACS=Acute coronary syndrome; CHF=Chronic heart failure; COPD=Chronic obstructive pulmonary disease; LISA=Longitudinell integrationsdatabas för sjukförsäkrings- och arbetsmarknadsstudier

Long-term cost-effectiveness analysis (study IV)

In study IV, the long-term cost-effectiveness of person-centred care provided to ACS patients was analysed. Thus, the cost-effectiveness calculations were performed for a time perspective longer than the ACS trial. A Markov-type model was constructed in order to project clinical outcomes beyond the reach of the clinical trial data. Calculations were performed for a two-year and a five-year perspective and included healthcare costs and indirect costs associated with sickness absenteeism from work and mortality. The health-economic model was constructed as follows. All patients start after an ACS-event state (remission). From that state, they can transit to one of three subsequent states: relapse, remission or death. The model accounts for two relapses and three remissions during the five-year period. Patients move between the states in 1 month cycles, based on transition probabilities estimated from the clinical trial data and from information collected from the scientific literature.

Methodological issues

In the subsequent sections, a summary account of the most important methodological issues encountered in the four studies is provided.

Missing data

Missing data occurred presumably at random in both randomized trials from which the data was collected. Challenges posed by missing data are more or less present in all empirical studies. The available methods for dealing with this problem include, for instance, imputation. Essentially, imputation involves replacing missing values with estimated values based on observed values [74, 116]. Imputation can be either single or multiple. Single imputation means that predicted values, obtained from one single calculation for each value, replace every position with missing data. However, single imputation methods do not account for uncertainty in the imputations. The multiple imputation approach seeks to mitigate this uncertainty by computing, instead of one single prediction, a series of predictions and corresponding multiple datasets [117]. The datasets are thereafter combined into one dataset to obtain the overall estimates. Multiple imputation by chained equations is a possible method of multiple imputation when several variables have missing values. The variables for which there are missing values are regressed on all analytically important variables in the data, using the

observations without missing data. The missing values are then replaced with predictions generated by the fitted equations. This procedure is then repeated for all other missing values [118]. Predictive mean matching (PMM) is one method for finding suitable values to impute. The method essentially imputes observed values from individuals similar to the missing case (called neighbours). The number of nearest neighbours is usually set somewhere between 3 and 10 [119]. In the case of missing baseline values, mean imputation (using values from both the treatment and the control arm), multiple imputation or exclusion of individuals from the analysis can be used [120, 121].

Multiple imputation by chained equations with PMM was used and missing baseline values were treated with the method of mean imputation in paper II and III. In addition, multiple imputation and bootstrapping (explained below) was performed simultaneously in order to infer the confidence interval for the cost-effectiveness ratio at the same time as accounting for missing values [122]. Imputation of missing EQ-5D values was performed on an index level [123]. For paper IV mean imputation for both missing baseline values and for the subsequent missing values was used.

Further, truncated data occurred due to patients dying during the trials. All deceased individuals were assigned a zero quality of life utility weight for all time periods after the time of death. Analyses including and excluding deceased patients were performed and presented in both paper II and paper III. Information from individuals under the age of 65 and that had survived throughout the trial was used to employ the model in paper IV.

Willingness to pay

When conducting cost-effectiveness analyses the results are usually presented in terms of the price for one additional quality-adjusted life year. This price is then usually compared to a given threshold, informal or formally announced by a governmental institute. In Sweden, there is an informal willingness-to-pay threshold of 500 000 SEK/QALY [124, 125]. This informal willingness-to-pay threshold was used in the cost-effectiveness studies in paper II, paper III and paper IV.

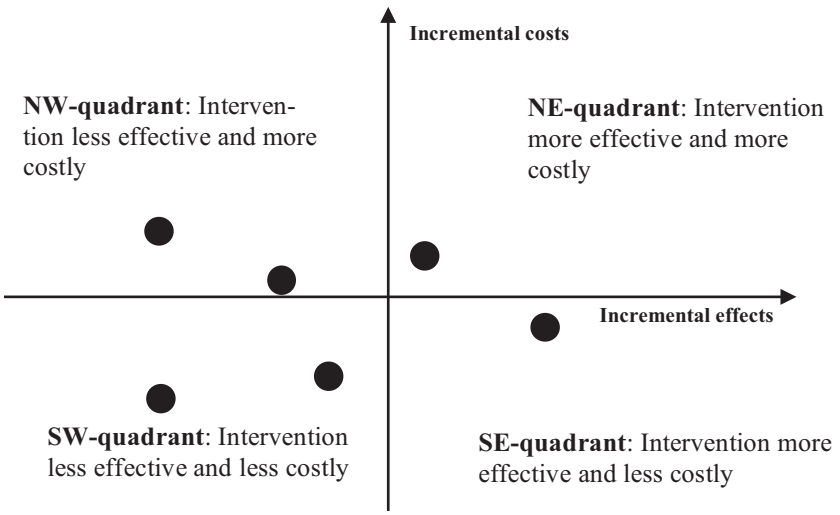
Uncertainty

Costs and health outcomes associated with particular health conditions and competing resource allocations in the healthcare sector cannot be measured perfectly.

In addition, these costs and outcomes are to a certain extent stochastic. Thus, uncertainty is an inherent feature in all health-economic evaluations [74]. Three main approaches exist to deal with this. First, in a deterministic sensitivity analysis variables used in the calculation of the cost-effectiveness measure are varied, one or several at a time, and the cost-effectiveness measure is recalculated using the alternative values on the variable(-s) [74]. Second, the objective in a probabilistic sensitivity analysis is to infer the uncertainty of the cost-effectiveness measure by recalculations using randomly drawn values of the variables beset with uncertainty. This is usually referred to as Monte Carlo simulation. Third, when primary data is available the method of bootstrapping may be applied, in which a random sample of the population is drawn and the cost-effectiveness measure calculated for this sample. Then the procedure is repeated for the entire data set (the analysed data is replaced after each iteration). The variance in the set of calculated cost-effectiveness measures is then used as a measure of the uncertainty of the cost-effectiveness measure [126, 127].

Since primary data was available, bootstrapping was used in study II, III and Monte Carlo simulation in study IV, where both primary data and data from the scientific literature was used.

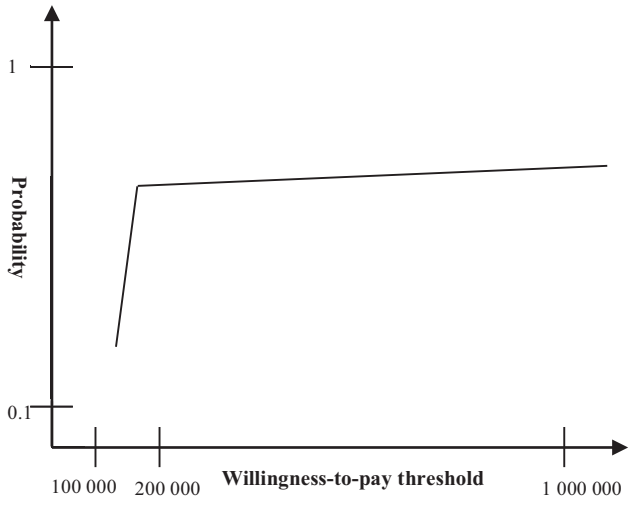
Figure 4. Illustration of a cost-effectiveness plane



NW = North-west; NE = North-east; SW = South-west; SE = South-east

In order to illustrate the uncertainty or variance of the cost-effectiveness measure, most often the incremental cost-effectiveness ratio, all incremental cost-effectiveness ratios attained from the Monte Carlo simulation or from the bootstrapping procedure are plotted in the cost-effectiveness (CE) plane [126, 127]. For example, the bootstrapping procedure executed with 1 000 sampling and replacements, produces 1 000 pairs of incremental effects and incremental costs that can be plotted in a diagram with incremental costs on the vertical axis and incremental effects on the horizontal axis (the CE plane). The CE plane is divided into four quadrants (see figure 4). Note, that the incremental cost-effectiveness ratio associated with each such pair is given by the slope of the straight line through the origin and point in the diagram. Point estimates of the incremental costs and effects may, in principle, end up in any of these quadrants. It is often the case that a new treatment alternative is both more costly and more efficient than the comparator alternative. In such cases the associated pair of incremental costs and effects is located in the first quadrant (NE). Similarly, if a new treatment is less costly and more effective the pair of incremental costs and effects would be located in the fourth quadrant (SE). In such cases, the new treatment is said to dominate the comparison treatment [128]. An illustration of the cost-effectiveness plane (CE-plane) is given in figure 4. A cost-effectiveness acceptability curve (CEAC) summarizes the uncertainty of an ICER and illustrates the probability of cost-effectiveness (how many of the incremental cost and incremental effect pairs fall below, and to the right of the line in the CE-plane) for different willingness-to-pay thresholds [129]. In figure 5 an illustration of the CEAC can be found.

Figure 5. Illustration of a cost-effectiveness acceptability curve



Discounting

In analyses where the time horizon is relatively long, costs accruing in the future are usually discounted to their present values. Analogously, future utility is worth less today. In both cases, present values can be calculated using the rate of interest and the rate of time preferences, respectively [75]. Typically, health-economic evaluations are concerned with time periods for which these considerations cannot be ignored. In the health-economic literature, costs and effects are usually discounting by annual rates ranging between 0 % and 6 %, for both costs and health effects [130]. In Sweden, the Dental and Pharmaceutical Benefits Agency (TLV) recommends a 3 % discount rate for both health effects and for costs followed by sensitivity analyses employing 0 % and 5 % discount rates [131].

A 3 % discount rate for both effects and costs in study IV was employed for the main analysis and 0 % and 5 % discount rates for effects and costs simultaneously or sequentially in the sensitivity analysis.

Intervention costs

Traditionally when conducting cost-effectiveness analyses the cost of the intervention itself is included in the cost estimates. In the case of person-centred care the inclusion of intervention costs is not straightforward. Person-centred care is more of a philosophy in care and a way of working for health personnel than it is an add-on healthcare approach to usual care. Because person-centred care is a philosophy and a way of approaching patients, it can be argued that giving person-centred care does not take longer time or more resources compared to giving usual care for a specific care contact. Hence, implementation of person-centred in healthcare practice would not incur any increased variable costs. There is a fixed cost when implementing person-centred care in practice which comprises of training healthcare professionals in person-centred care. The attained knowledge from training can be applied on a great number of patients over time and therefore the fixed costs of implementing person-centred care can be expected to approach zero. Therefore, no intervention costs were added to the cost-effectiveness analyses in paper II and paper III. The aim of paper IV was to create a model which could then be used for other interventions for patients with ACS. Because other interventions might have both a fixed cost and a variable cost, these aspects were added to the Markov model as an alternative.

Choice of outcomes

Person-centred care has been evaluated using a range of different outcome measures and there is no consensus concerning the appropriateness of these measures in studies on the effects of person-centred care. Below are descriptions of the health-related outcomes used in our studies.

Health-related quality of life

Health-related quality of life, measured by the EQ-5D-3L questionnaire, was used in all four studies [132]. The EQ-5D-3L questionnaire consists of five questions with three dimensions to each question and deals with level of mobility, anxiety, hygiene, activity and pain. The EQ-5D questionnaire is frequently used in economic evaluations in order to measure health-related quality of life [133]. The scores are compiled and translated into QALYs, which is the most common outcome measure used in health-economic evaluations [133]. This translation assigns values to different health states using value sets. Value sets can either be hypothetical, meaning people from the general public set values to different health states without themselves being in the actual states, or experience based. An experience based value set is based on individuals who are in the actual health states. Advocates for a hypothetical value set claim that since healthcare is mostly financed by the society collectively, the society should also give values to health states. In comparison, advocates for the experience based value set argue that it is indeed the persons experiencing the health states who can value them the best. In 2013, an experience based value set was introduced in Sweden and is today recommended by the Dental and Pharmaceutical Benefits Agency when performing studies with QALYs [131, 134, 135]. In study I the UK tariff was used to translate EQ-5D to QALYs and in the remaining studies the Swedish, experience based, tariff was used.

General self-efficacy

General self-efficacy (GSE) is a measure of a person's belief in his or herself and the abilities to cope with certain problems or setbacks, as well as the ability to recognize symptoms and how to deal with these. GSE is measured using the General Self-Efficacy Scale which comprises of 10 questions with 4 dimensions and the total score for the questionnaire is between 10 and 40 points (40 points being "most self-efficient") [136]. General self-efficacy was used as a health-related outcome measure in study I as the measure is associated with increased motivation to cope with the disease at hand by targeting internal confidence

[137]. Therefore, the measure is closely related to self-management and person-centred care and an important health-related outcome to consider when analysing a person-centred care approach [12, 114].

Other outcomes

In study I the effects of person-centred care were measured using level of physical activity and return to work, in addition to the two previously mentioned measurements. Desire of returning to work or to previous leisure activities are goals expressed by patients with ACS and because person-centred care aims at shared-decision making and goal achievement the two former measurements were chosen as effect measures in the evaluation of the intervention [138]. The self-reported level of physical activity was measured using the Grimby Scale [139]. The scale consists of one question regarding physical activity during leisure time with four possible levels to choose from, ranging from physically inactive to regular hard training for competitive sports. Self-reported return to work was measured in a dichotomous variable; 1 if the individual had returned to work and 0 otherwise.

Ethical considerations

The two clinical trials from which the data used in this thesis was collected and the access to healthcare registers have been approved by the Ethics committee (Dnr 275-11; Dnr 687-14; and Dnr T445-16; Dnr T812-17). The patients involved in the studies have given their written consent and have been informed that the individual information collected will be used in research and will be published in aggregated form. The participants have also been informed that additional information will be collected from different health registers. No commitments as to direct benefits accruing to the patients involved in these studies have been made. The knowledge generated by health-related research in general is more likely to benefit future than current patients and, in the case of cost-effectiveness research, the effectiveness of future public and private spending, rather than current spending. The respondents' privacy is protected by preventing unauthorized access to the data (personal identification numbers and responses to questionnaires are stored in a locked room, and all individual information is encrypted when sent to the register-holding authorities in order to complement the data collected within the trials).

Results

In this section, the results for the two person-centred care trials are presented separately. In table 2 the results from each study are summarized.

ACS

Below, the results from the ACS trial are presented. The results are divided into health-related outcomes and cost-effectiveness.

Health-related outcomes

The patients in the person-centred care group had statistically significant improvements in the general self-efficacy score compared to the usual care group when measured 6 months after randomization to the trial. The positive and significant effect on general self-efficacy appeared in the analyses after controlling for all explanatory variables in the regression analyses. The positive difference in physical activity between baseline and follow-up at 6 months, measured by the Grimby Scale, was larger for the person-centred care group compared to the usual care group. More patients in the intervention group than the control group reported that they had returned to work 6 months after randomization. The intervention effect on return to work and physical activity was statistically non-significant in all regression analyses. The EQ-5D score, measuring health-related quality of life in study I, at one year follow-up, was higher for the intervention group compared to the control group but was not statistically significant. The positive non-significant effect was only present when all explanatory variables were added to the analyses, otherwise the effect of the intervention on health was negative and non-significant.

Table 2. Summary of results divided by study

	Study I	Study II	Study III	Study IV
Effects of person-centred care	Significant and beneficial effects on general self-efficacy	Cost-effective and dominating alternative compared to usual care	Cost-effective and dominating alternative compared to usual care	Cost-effective and dominating alternative compared to usual care after 2 and 5 years
Type of patients	For entire study population	For patients under 65 years	Both for entire study population and without deceased patients	For patients under 65 years

Short-term cost-effectiveness

The short-term cost-effectiveness of person-centred care for patients with ACS was analysed for the entire study population as well as for subgroups. Overall, the results were ambiguous. In what follows, a more detailed description of the results is provided.

One exclusion criteria for the ACS-trial was estimated survival under 1 year at randomization. Nevertheless, 7 patients died during the first year after randomization, 5 in the intervention group and 2 in the control group. These deaths were assumed to be random and therefore the deceased patients were excluded from the main analysis and subgroup analyses. However, sensitivity analysis was performed for the entire study population of 199 patients (94 in the intervention group and 105 in the control group). The analysis was employed from a societal perspective with all direct and indirect costs included and results showed a likelihood of cost-effectiveness of person-centred care for the entire study population of 50 % for the willingness-to-pay threshold of 500 000 SEK/QALY. For the main cost-effectiveness analysis, the patients that had deceased during the first year after randomization were excluded. The analysis was then conducted on 192 patients, 87 in the intervention group and 103 in the control group. Results, after comparing costs and effects between the intervention group and the control group, showed that person-centred care was both less effective and less costly compared to usual care. Furthermore, the likelihood that person-centred care was cost-effective compared to usual care was 55 % for a 500 000 SEK/QALY threshold.

The majority of people in Sweden exit the workforce at the age of 65. Hence, productivity losses are likely to be of larger importance when considering patients under the age of 65. Therefore, a separate analysis was conducted for patients under the age of 65 and 65 years and older (excluding deceased patients in both cases) to investigate whether the cost-effectiveness of person-centred care would differ between the two groups. The older patient group consisted of 75 patients in total, 34 in the intervention group and 41 in the comparison group. For patients at the age of 65 and over, person-centred care resulted in larger costs and lower effectiveness compared to the group receiving usual care, i.e. usual care dominated person-centred care. The younger patient group consisted of 117 patients, 55 in the intervention group and 62 in the control group. The person-centred care intervention was both more effective and less costly compared to usual care for patients under the age of 65 years. Sensitivity analyses showed a likelihood of person-centred care being cost-effective compared to usual care of

89 % when utilising both indirect and direct costs and a likelihood of 86 % when employing only direct costs, for a willingness-to-pay of 500 000 SEK/QALY. Hence, person-centred care dominated usual care for the younger group, both when employing a societal perspective and a healthcare perspective.

Long-term cost-effectiveness

The long-term cost-effectiveness of person-centred care for patients with ACS under the age of 65 was analysed using a Markov-type model. Person-centred care was found to be the dominating alternative compared to usual care. Below, the results from paper IV are presented in more detail.

For the base-case analysis, data from the person-centred care intervention was used to create inputs for the developed Markov-type model. Person-centred care was found to dominate usual care, resulting in both less costs and more effects, when using a two-year time-perspective and including all costs. When excluding productivity losses due to mortality and thereafter indirect costs due to sick leave, person-centred care was still more effective and less costly compared to usual care after 2 years. Conducting the same analyses, only for a five-year time-perspective, the results showed that person-centred care was still more effective and less costly compared to usual care. Removing productivity losses due to mortality and thereafter employing a healthcare perspective resulted in quantitatively similar results. Sensitivity analyses conducted with the Monte Carlo approach resulted in 1 000 pairs of ICERs (based on 1 000 pairs of incremental effects and 1 000 pairs of incremental costs) which were analysed to obtain the likelihood of cost-effectiveness. The likelihood of person-centred care being cost-effective compared to usual care for a two-year time-perspective was estimated between 80 % and 98 % and the corresponding likelihood for a five-year time-perspective between 75 % and 90 %, both with a willingness-to-pay threshold of 500 000 SEK/QALY. Calculating the cost-effectiveness while simultaneously or sequentially employing a 5 % and a 0 % discount rate on costs and effects only altered the results slightly.

Scenario analysis was performed on inputs that were believed to be impacted by healthcare interventions designed for patients with ACS. Inputs were allowed to vary in order to produce cost-effectiveness ratios on both sides of the willingness-to-pay threshold of 500 000 SEK/QALY. All inputs were chosen from the group receiving person-centred care. In short, the monthly risk of first relapse (during the first 2 years after the initial ACS event) could increase from 0.925 % (base-case) to around 1.13 % and still result in a cost-effective intervention. Fur-

ther, the risk of a second relapse (during the first 2 years after the initial ACS event) could increase from 0.56 % (base-case) to 5.86 %; indirect costs due to short-term sickness absenteeism from SEK 8 250 (base-case) to SEK 342 290 per month; healthcare costs associated with being in remission, from SEK 2 223 (base-case) to SEK 2 741 per month; and healthcare costs associated with relapse, from SEK 39 824 (base-case) to SEK 107 302 per month.

COPD/CHF

Below, the results from the COPD/CHF trial are presented. Only short-term cost-effectiveness analysis was performed for the COPD/CHF trial.

Short-term cost-effectiveness

For the main cost-effectiveness analysis, the entire study population of 221 patients (103 in the intervention group and 118 in the control group) was analysed. The incremental effect was positive and the incremental cost was negative, meaning the person-centred care intervention was both more effective and less costly compared to usual care. The probabilistic sensitivity analysis showed that the likelihood of person-centred care being cost-effective compared to usual care for a 500 000 SEK/QALY threshold was 95 %. In the subgroup analysis, the patients that had deceased during the first 6 months after randomization were excluded. As a result, the study population consisted of 184 patients, 87 in the person-centred care group and 97 in the usual care group. Still, the person-centred care intervention was more effective and less costly, i.e. person-centred care dominated usual care when both including the entire study population and when excluding the deceased patients. Conducting sensitivity analyses resulted in a likelihood of person-centred care being cost-effective compared to usual care of 87 % for a threshold of 500 000 SEK/QALY, when excluding deceased patients.

Discussion

This thesis contributes to the small but emerging literature on the effects and cost-effectiveness of person-centred care. In summary, the results obtained in the four studies suggest that person-centred care has positive effects on health-related outcomes and is the cost-effective alternative when compared to usual care, both in a short and in a long-term perspective. Of course, these results pertain to specific contexts, in particular, different therapeutic areas. Future research should expand on these efforts in order to further improve the applicable knowledge on the cost-effectiveness of person-centred care.

The results obtained in the four studies suggest (1) that person-centred care improves health-related quality of life, and (2) that person-centred care is cost-effective compared to usual care when provided to patients with ACS (below 65 years of age) and COPD and/or CHF.

In what follows some of the methodological issues encountered in the four studies will be addressed and discussed. The section ends with a general discussion and policy implications.

Methodological issues

Costs and cost-effectiveness

Register data was employed in order to calculate the healthcare costs associated with person-centred care (and usual care) in studies II, III and IV. Observed healthcare utilization was transformed to monetary costs using the diagnosis related groups (DRG) system. Although this is an established method, one needs to be aware of its potential drawback: cost calculations are performed using the clustering of individuals that the DRG system entails. That is, patients receiving the same DRG points may, in fact, differ somewhat as regards to actual healthcare received. This discrepancy is likely to be small, or even insignificant, since measurement errors aggregated over all observations can be assumed to, at least partly, cancel out. In principle, information about individual healthcare

resource utilization is available from medical journals. However, collecting this information is impractical.

Indirect costs – productivity losses due to sickness absenteeism – were included in studies II and IV. The costs associated with absenteeism were estimated employing the human-capital approach, which prescribes measuring the costs as the market value of lost production. In a perfect competition situation, this would be unproblematic since observed incomes would be a perfect measure of the value of production and since information would be perfect. Of course, all real situations diverge more or less from this ideal. Aggregate mean wages, as reported by Statistics Sweden, were used as estimates of the value of production, since information about individual wages were not possible to collect within the trials. The aggregate figures may diverge more or less from the true wages received by the respondents in the clinical trials. Again, the argument that measurement errors may to some extent cancel out and leave the overall measure of the value of productivity losses close to its true value applies.

An additional factor potentially affected by person-centred care, which was not considered in any of the studies, is informal care. It was demonstrated in study I that person-centred care influences self-efficacy positively. In light of this result it seems not too farfetched to assume that person-centred care reduces the need for and the utilization of informal care. Thus, including also informal caregivers' support, and the indirect costs thereby evoked, would further strengthen the finding that person-centred care is cost-effective compared to usual care.

Person-centred care was assumed to have a zero cost in all cost-effectiveness calculations in studies II – IV. The rationale for this is that the implementation of person-centred care only requires a fixed initial cost (a workshop and a number of booster sessions for the healthcare personnel). This human-capital investment is then used on a large number of patients over time and, hence, driving the per-patient cost towards zero. There are (at least) two caveats to note. First, the personnel's knowledge on how to provide person-centred care may depreciate over time. However, this argument seems to be valid only when the acquired knowledge is not put to use. In reality it is far more likely that the capability of providing person-centred care will improve over time due to on-the-job training. Second, when person-centred care is delivered as an add-on to usual care there will, in principle, be a variable cost involved. This was the case in study III. However, two recent studies involving the same add-on component found that the additional cost of providing person-centred care is insignificant compared to other healthcare costs [48, 115].

Data collection

Empirical research always involves dealing with various data issues. In particular, the process of collecting data is not perfect and, therefore, some observations may be missing or truncated in the resulting dataset. Missing or truncated observations occurred for two reasons in both datasets that were used in the studies: incomplete questionnaires and deaths occurring during the clinical trials. These events were assumed to occur randomly and, hence, to have no effect on the true effectiveness and cost-effectiveness of person-centred care. Missing data from incomplete questionnaires were dealt with by multiple imputation of the missing observations. In contrast, the effect of truncated data due to deaths was inferred by performing analyses with and without those patients, where included deceased patients were assigned a zero quality of life and zero costs after death. Of course, this procedure can only to a limited extent account for the information that would have been present in the data had no patient died. The imputation of missing data was performed employing predictive mean matching, since this method deals with, the usually, non-normal distribution of health-related quality of life weights. Multiple imputation (instead of, for example, single imputation) was used to deal with the uncertainty surrounding missing data and the method replaces the missing values with a set of plausible predicted values instead of one single value [121]. The non-response analysis suggests that missing values occurred unsystematically and, hence, are not likely to bias the results obtained in the studies.

Study population

A further caveat concerns the patients that were excluded from the clinical trials. The exclusion criteria adopted in the two clinical trials resulted in the exclusion of very old patients (over the age of 75 for the ACS trial), and patients with an estimated survival less than one year (both trials). As a result, the study populations in the clinical trials do not perfectly correspond to the targeted populations when person-centred care is implemented in clinical practice. Studies performed on the effects of patient-centred care provided to patients with other health conditions than ACS have found that patient-centred care increases health-related quality of life, also for very old patients [59]. Thus, to the extent that the results are valid also for person-centred care provided to patients with ACS, complementary cost-effectiveness calculations for elderly patients are feasible using this information.

General discussion

Finally, I want to address the question of to what extent the aims of the thesis were met by the studies actually performed. The overall aims of this thesis were (a) to improve and extend available and applicable knowledge of the cost-effectiveness of particular person-centred care interventions and (b) to improve and extend the methods for performing health-economic evaluations in the area of person-centred care, by exploring the usefulness of health-economic methods and various outcome measures in different disease and institutional contexts.

Clearly, all studies improve and extend available knowledge concerning the effectiveness and the cost-effectiveness of person-centred care. Moreover, the cost-effectiveness results are directly applicable to clinical situations. The question of the general validity and, hence, the applicability, of the results is, as always, a delicate matter. Intuitively, it seems plausible that the results cannot be directly transferred to situations involving other health conditions than the ones studied. This reinforces the conclusion above that future studies need to examine health conditions hitherto not studied. In contrast, it seems more plausible that the results can be assumed to be valid for healthcare settings other than the ones in which the clinical trials, from which the data was collected, were performed.

To improve and extend the methods for performing health-economic evaluations in the area of person-centred care is obviously an ambitious goal. The performed studies do achieve this to some extent. Study I gives some guidance as to the applicability of different outcome measures and, hence, also provides guidance on how to specify cost-effectiveness calculations in the particular case of person-centred care. There is a possibility that particular empirical measures do not capture or are not sensitive to treatment effects achieved by a specific intervention. In order to facilitate cost-effectiveness comparisons between different therapeutical areas the preferred outcome measure to be used is a utility-based measure. The results in study I demonstrate that the EQ-5D measure is, to some extent, sensitive to the effects achieved by person-centred care. Moreover, study I may serve as a basis for extending the health-economic evaluation methodological toolbox by taking individual capabilities into account using the demand-for-health framework as a point of departure.

Future research should further study the applicability of different measures to capture the effects of person-centred care. The various outcome measures employed in studies of person-centred care, and other similar complex interventions designed with the patient/person in mind, include health-related quality of life

[24, 36, 59-62, 64], ADL [6, 22, 57], costs [6, 48] and length of hospital stay [54, 140]. There is no established practise as to what particular measures that are most appropriate in each particular situation [10, 24]. Frequently used outcome measures include general self-efficacy and more disease specific measures of self-efficacy. These measures seem to comprise dimensions which are affected by these types of interventions [27, 30, 52, 53, 56-58, 114, 141-143]. A possible explanation for this is that the objectives of person- and patient-centred interventions typically are to increase a person's confidence in symptom management and in the ability to achieve personal goals, and to enhance the ability to cope with the disease, which are domains that the general self-efficacy measure aims to capture [113, 144, 145]. For reasons already stated, preferred outcome measures to be used in health-economic evaluations are based on utilities. Only health-related quality of life among the aforementioned outcome measures may fulfil this requirement.

The person-centred care interventions studied in this thesis are closely related to the capability approach by Sen [146]. Sen emphasizes the importance of a person's capabilities rather than his or her preferences. In other words, Sen's capability approach puts an emphasis on what an individual can do rather than on what he or she would like to do. Person-centred care aims to guide the patient in finding personal resources and strengths, but also weaknesses and barriers, and to set and reach goals. This bears a close resemblance to improving capabilities. However, typically, traditional health-economic outcome measures do not capture individual capabilities and, hence, to the extent that person-centred care based interventions enhance individual capabilities the effects of such interventions are poorly captured by these measures. Thus, there is a need for the development of outcome measures that more comprehensively captures the effects of person-centred care, assuming that the current utility-based measures used within the extra-welfarist approach are imperfect in this respect. That this is the case seems likely since the instruments used for collecting utility weights have functioning rather than capability in focus [147]. One existing measure that potentially improves on this situation is the Investigating Choice Experiments Capability Measure (ICECAP), which is grounded on Sen's theories on capabilities and functioning [148, 149]. Attempts to construct ICECAP indexes pertaining to adults have been made and will potentially be useful in the evaluation of health and social care interventions [148, 149]. The correlation between ICECAP and EQ-5D is low and, hence, combining the ICECAP and the EQ-5D measures would provide even more comprehensive information on the effects of person-centred care [150]. There are, however, other methodological issues involved in applying the capability approach. For instance, the objective of a social planner

when allocating resources is not as straightforward as in traditional cost-effectiveness analyses. One alternative would be to allocate resources in order to obtain a minimum level of capability to as many people as possible [151].

Further, a broader definition of health including the ability to adapt and to self-manage [152], may be useful for capturing the effects of person-centred care. Payne and co-authors [153] made an important point by stating that it is essential to identify a balance between health and non-health gains produced by an intervention that aims at, not curing patients, but offering them an adjustment and hope for the future for incurable diseases that are not possible to treat with surgical or medical interventions. Person-centred care aims at achieving this, but an outcome measure which comprehensively captures these effects and is appropriate for health-economic evaluations still remains to be developed.

Policy implications

The overview of empirical evidence regarding the effectiveness and the cost-effectiveness of person-centred care presented in the background section suggests that person-centred care is both an effective and a cost-effective alternative, compared to usual care, at least for some health conditions. The strength of the general conclusions concerning the effects and the cost-effectiveness of person-centred care are conditioned, however, on the assumption that available empirical evidence pertaining to other types of similar, but not identical, interventions have some bearing for person-centred care based interventions [13]. The summary provided in the background section, reveals that there are considerable gaps in the knowledge about the effectiveness and the cost-effectiveness of person-centred care. In particular, future research needs to investigate the effects of person-centred care for a broader range of health conditions. As for the cost-effectiveness of person-centred care, there seems to be a lack of evidence beyond what available clinical data would permit. Moreover, future clinical research should involve health-economic considerations already at the planning and the design of trials, since the need for information required by effectiveness studies and studies concerned with cost-effectiveness is not entirely overlapping.

The three studies on cost-effectiveness largely indicate that person-centred care is cost-effective compared to usual care for the health conditions under consideration. Two main questions are important to address when it comes to the policy

implications that can be drawn from these results, i.e., when to implement person-centred care in the clinical healthcare practise: the validity of the results for patient groups and health conditions other than the groups and conditions included in the clinical trials. In the absence of further research providing solid cost-effectiveness results for particular patient groups the best option seems to be to adopt a heuristic approach based on available cost-effectiveness results and on the intuition of the healthcare personnel. Of course, cost-effectiveness is not the only principle that should guide decisions on how to allocate healthcare resources. In the Swedish context, there are three governing principles: the human dignity principle, the needs-solidarity principle, and the cost-effectiveness principle. This order is hierarchical and, hence, the cost-effectiveness principle should be considered last.

Conclusions

In this thesis, the effects and the cost-effectiveness of person-centred care was examined employing primary data from two person-centred care clinical trials. Person-centred care was found to increase self-efficacy significantly (study I); to be cost-effective after 1 year for patients with ACS under the age of 65 (study II); to be cost-effective after 6 months for patients with COPD and/or CHF (study III); and to be cost-effective for a two-year and a five-year time perspective for patients under the age of 65 with ACS (study IV).

Carefully formulated, the results obtained in this thesis suggest that person-centred care should be considered as an alternative to usual care, particularly for patients with chronic diseases such as ACS, COPD and CHF.

This thesis contributes to the growing but still scarce knowledge concerning the effects and the cost-effectiveness of person-centered care. The results largely corroborate previous findings concerning the effects and the cost-effectiveness of person-centred care. Even though person-centred care has already been implemented in several healthcare practices in Sweden [154], the results obtained and presented in this thesis provide additional guidance for future implementations of person-centred care.

Future developments

Perhaps the most important directions for future developments as regards to the cost-effectiveness of person-centred care is to extend the perspective to other health conditions and contexts than those studied here. Although ACS, COPD and CHF, are common diseases, several other health conditions need to be examined in order to achieve an "appropriate" level of knowledge. The interventions examined in this thesis pertain to chronic diseases and are delivered through face-to-face interactions and through phone-calls. Obvious developments would be to examine the effects of person-centred care when delivered in other contexts and to patients with health conditions that are not chronic.

In addition, and as hinted above, the possibly most important issue concerns the development of outcome measures that more fully capture the effects of person-centred care. A potential way forward involves interviewing patients who have received person-centred care and investigate which dimensions they considered most important and what, if any, aspects differed from the care that they usually receive. Using such information may prove useful for designing improved outcome measures.

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Appendix

The scientific literature on the effects and the cost-effectiveness (both short and long-term) of person- and patient-centred care, self-management and disease management programs is summarized in Table A1. The summary is confined to peer-reviewed journal articles. Searches were performed in Pubmed, CINAHL and Scopus using the search phrases:

(((“person-centred care”) OR (“patient-centred care”) OR (self-management)) AND ((“cost-effectiveness”) OR (“quality of life”) OR (self-efficacy) OR (“economic evaluation”)))

Also, with the search phrases:

(((“person-centred care”) OR (“patient-centred care”) OR (self-management)) AND ((“Markov”) OR (“extrapolation”))).

The searches were executed during February 2019. Studies were included if they evaluated the effects or cost-effectiveness of person-centred care, patient-centred care, self-management programs or disease management programs that had something in common with the structured approach of GPCC interventions and if outcomes used were health-related quality of life or self-efficacy. Studies were excluded if they were not written in English or not peer-reviewed. This review of the literature does not fulfill the requirements of a full systematic literature review. Still, the summary gives an approximate appreciation of the current literature extant in this field of research.

Table A1: Summary of previous research on the effects of patient-centred care and person-centred care, presented by disease and outcome measures used

	Cost-effectiveness	Health-related quality of life	Self-efficacy	Disease specific/general quality of life or health status	Reduction in hospital stay	Reduction in costs
ACS	*Person-centred care cost-effective (Pirhonen et al., 2019)*	*Positive, non-significant effects of person-centred care (Pirhonen et al., 2016)*	*Significant effects of person-centred care (Fors et al., 2015ab, Pirhonen et al., 2016)*			
Cancer	*Person-centred care dominates usual care (Gyllensten et al., 2018)* and collaborative care planning cost-effective (Sandsund et al., 2017)	*Improvements after person-centred care (Olesen et al., 2016)*				*Decreased costs of person-centred care (Gyllensten et al., 2018)*
Chronic heart failure	*Person-centred care less costly compared to standard care (Hansson et al., 2016) and cost-effective (Pirhonen et al., 2019)* and disease management program cost-effective (Bocchi et al., 2018)	Significant effects of person-centred care (Sahlen et al., 2016) and patient-centered care (Kane et al., 2015).	Improvement from person-centred care (Ulin et al.., 2015)	No effects of patient-centred care (Bekelman et al., 2015)		*Decreased costs of person-centred care (Hansson et al., 2016)*
Chronic kidney disease		Improvements from self-management programs (Lin et al., 2017)	Improvement from self-management programs (Havas et al., 2018; Lin et al., 2017)			

Chronic liver failure	Disease management program cost-effective (Wiggs et al., 2018)					
COPD	*Person-centred care cost-effective (Pirhonen et al., 2019)*					
Dementia		Improvements after patient- and person-centred care (Kim et al., 2017; Tay et al., 2018; Ballard et al., 2018; Sjögren et al., 2013)				
Depression	Self-management intervention not cost-effective for anxiety, depression and somatoform disorders (Grochtdreis et al., 2018)	Improvements after self-management program (Turner et al., 2015).	Improvements after nurse-led self-management support for patients with anxiety, depression and somatic symptoms (Zimmerman et al., 2016)			
Diabetes			Greater self-efficacy from nurse follow-up calls (Piette et al., 2000)	No significant effects of patient-centred care (Glasgow et al., 2005)	No significant effects of self-management program (Lorig et al., 2010)	
Hip fracture and hip arthroplasty	*ICP treatment less expensive and more effective than standard care (Olsson et al., 2009)*				*Reduction for hip fracture and hip arthroplasty patients after person-centred care intervention (Olsson et al., 2006; Olsson et al., 2014)*	
HIV				Significant effects of patient-centred care (Gustafson et al., 1999)		

Inflammatory bowel disease	Patient-centred care cost-effective (Richardson et al., 2006)					
Intellectual disabilities		Improvements after person-centred planning (Ratti et al., 2016)				
Pancreatic disease	Cost-effective management as a result of patient-centred care (Pendharkar and Petrov, 2015)					
Psoriasis	Patient-centred care less costly but as effective as standard follow-up treatment (Parsi et al., 2012)			No significant effect between on-line patient-centred follow-up versus standard care (Chambers et al., 2012)		
Stroke		Self-management programs increase health-related quality of life (Fryer et al., 2016).	Improved self-efficacy as a result of self-management programs (Fryer et al., 2016; Lo et al., 2018)) and patient-centred care (Chen et al., 2018)			

Studies on chronic conditions		No significant effects from self-management program (Kennedy et al., 2013), nurse-led tele-health intervention (Looman et al., 2018), personalized care planning (Coulter et al., 2015) or patient-centred care (Salisbury et al., 2018)	Significant effects from patient-centred care (Rutten et al., 2016)	Disease management led to improvements (Yukawa et al., 2010; Lorig et al., 1999). Significant effects of patient involvement (Kaplan et al., 1989). Improvements from a collaborative care model (Katon et al., 2010) and self-management program (Reed et al., 2018)	Reduction from a self-management program (Lorig et al., 1999)	

*The table is based on a table from study 1 (Pirhonen et al., 2017) and has been updated with additional articles. Only articles using outcomes reflecting quality of life, cost-effectiveness or reduction in costs are presented and articles somewhat close to patient-centred or person-centred care. Studies that examine GPCC interventions are highlighted in *italics*.