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Empirical Evidence of the Impact of Integrated Care Interventions for Persons with Multi-Morbidity using Multi-Criteria Decision Analysis

Maaike Hoedemakers

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Empirical Evidence Of The Impact Of Integrated Care Interventions
For Persons With Multi-Morbidity Using Multi-Criteria Decision Analysis

MCDA:
Expliciet, Uitgebreid, Transparant

Empirisch bewijs van de impact van integrale zorg
voor mensen met multi-morbiditeit met behulp van Multi-Criteria Decision Analysis

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Set your sights upon the heights
Don’t be a mediocrity
Don’t just wait and trust to fate
And say, that’s how it’s meant to be
It’s up to you how far you go
If you don’t try, you’ll never know
And so my lad as I’ve explained
Nothing ventured, nothing gained

- Merlin *(The Sword in the Stone – 1963)*

Voor Nolan en Sverre

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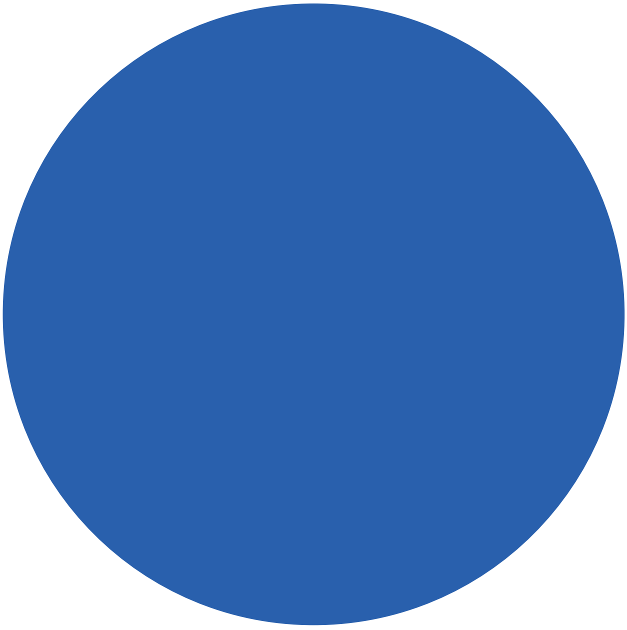
Chapter 1

General introduction



THE BURDEN OF MULTI-MORBIDITY

The increasing prevalence of multi-morbidity in Europe – over 50 million persons in Europe have more than one chronic condition – affects both individuals and society and constitutes a challenge for health systems.[1] On average, almost one third of people aged 15 years and over across 27 OECD countries suffer from multi-morbidity.[2] In the Netherlands, 27% of people 15 years and older, and 50% of people 65 years and older reported living with multi-morbidity. The prevalence of multi-morbidity is expected to further increase as a result of population ageing, advanced medical technology, and increased exposure to risk factors.[3,4] People with multi-morbidity are at risk of poorer health outcomes, decreased health-related quality of life, and higher mortality than persons without a chronic condition or just one condition.[5-11] Furthermore, multi-morbidity places a high burden on human and financial resources. People with multi-morbidity are prone to working less hours or exit the labour market early.[12] Productivity losses could also occur in informal caregivers as the cumulative load of working and taking care of a loved one is considered challenging, leading to reduced work hours in informal caregivers.[13,14] Moreover, multi-morbidity leads to higher healthcare expenditure caused by greater healthcare utilisation, an increased risk of unplanned hospital admissions, and longer hospital admissions.[15-17]

The Organisation for Economic Co-operation and Development (OECD) expects the proportion of older people to be increasing due to a combination of declining fertility rates and increased life expectancies.[18] While the percentage of the population aged over 65 years is expected to increase from 17% in 2015 to 28% in 2050 across OECD countries, the proportional increase of people older than 80 years is projected to be even larger, i.e., from 3% in 2015 to 14% in 2050.[19] Moreover, the ratio of working age people (15-64 years) per person aged older than 65 is forecasted to be halved from 4.2 to 2.1 by 2050.[20] Hence, the rising healthcare expenditures must be borne by less working people through income taxes and/or employers' contributions to health insurance premiums. The economic burden is substantial and healthcare expenditure in the Netherlands is already among the highest in Europe, both as percentage of GDP (NL: 10.0% *vs* European Union (EU) average: 8.3%) and per capita (NL: 3,908 EUR PPP *vs* EU average: 2,572 EUR PPP).[18,21] Focusing on the Netherlands, life expectancy at 65 years increased from 18.4 years in 2005 to 20.3 years in 2019. However, this does not necessarily mean that these extra years are spent in good health, as the overall time spent in good health (measured as the number of years that one can expect to live free of disability) has been declining from 59.0% to 48.7% over these years.[22,23] Subsequently, population ageing has led to an increased demand for long-term care, which is a key driver of expenditure. Compared to EU countries, the Netherlands spends the highest percentage of its total healthcare budget on health-related long-term care, i.e., covering 27% of total healthcare spending.[18]

UNMET NEEDS OF FRAIL ELDERLY

The demographic and epidemiological trends described above also contribute to an increasing number of frail older persons with complex care needs, as there is a strong association between ageing, multi-morbidity, and frailty.[24- 26] Studies show that the prevalence of multi-morbidity among frail persons was 72%.[25,27] Frailty can be defined as a clinical syndrome of increased vulnerability due to ageing-associated physical and cognitive decline that challenges people to cope with acute stressors (e.g., a bladder infection) and that carries an increased risk for poor health outcomes, such as falls, hospitalisation, and mortality.[28] With a focus on the physical aspect of frailty, this was further operationalised by Fried and colleagues (2001), who defined frailty as the presence of as least three of the following criteria: unintentional weight loss, self-reported exhaustion, weakness, slow walking speed, and low physical activity.[29]

Frail older people have complex care needs that transcend the healthcare domain. As a result, frail elderly often receive care from both health- and social care providers, in primary and secondary care, and chronic and acute care. Thus, a multitude of care providers is involved.[30] Therefore, frail older people with complex care needs are at risk for care that is fragmented, duplicated, and directed at acute disease, which interferes with delivering efficient and effective chronic care.[31] Fragmentation of care occurs when patients are being transferred to other care providers or institutions without complete information about the patient’s conditions, medical history, or previously described medications. Patients are then faced with unnecessary risks, waiting times, and duplication of tests or treatments, which also brings along avoidable healthcare costs.[32] Furthermore, current care delivery for persons with multi-morbidity is hampered by single-disease clinical guidelines, as a result of which patients are at risk of unforeseen treatment interactions, conflicting treatment goals between multiple care providers, and overestimated self-management capacities.[33,34] Besides a negative impact on healthcare costs, this could adversely affect patient outcomes and experience with care.[35] Another criticism on the health system is that care delivery is often reactive, in that it primarily starts after the patient actively seeks help from their general practitioner (GP). Although the health system has evolved over the last decade, there is still room for improvement to gear towards delivering care in a more proactive and preventive manner.[36]

INTEGRATED CARE

In response to the increasing number of frail older people with complex care needs and the inadequate response of the healthcare system, reforms have taken place to move towards more integrated care. There are many typologies of integrated care and the focus of the definition shifts depending on the perspective from which it is defined. The World Health Organisation (WHO) takes a health system perspective and defines integrated care as: *"an approach to strengthen people-centred health systems through the promotion of the comprehensive delivery of quality services across the life-course, designed according to the multidimensional needs of the population and the individual and delivered by a coordinated multidisciplinary team of providers working across settings and levels of care."*[37] Integrated care is aiming to improve patient outcomes, experience with care, and to decrease health- and social care costs.[38] This requires new models of care that put person-centred care and care coordination central. Such innovative care models often link the healthcare sector to the social care and welfare sector, the mental healthcare sector, and the health promotion and prevention sector, hence requiring multidisciplinary collaboration across a wide range of disciplines.

Integrated care is also expected to lead to efficiency gains. This increased interest in efficiency can be observed across the whole spectrum of care delivery, since not only health insurers are paying for care of patients with complex care needs. Due to healthcare reforms in The Netherlands, municipalities take on the role of payer for domestic (home) care and social care support. Care groups (i.e., associations of primary care providers that develop chronic care programmes and support the provision of such programmes) are interested in efficiency gains as they are negotiating with healthcare insurers about payments for integrated care, mostly through bundled payments.[39,40] Hence, there is great interest in the (cost-) effectiveness of integrated care. As a euro can only be spend once, policy makers must decide whether to invest in integrated care, as then there will be less budget available for existing interventions or other innovations. Hence, we need robust information on (cost-) effectiveness to ensure the prioritisation of interventions that deliver the most value for money. However, recent studies found little solid evidence of the improvement of patient outcomes or the cost-effectiveness of these new models of care.[38,41-43] Studies showed that the methodological quality of economic evaluations in integrated care was low, which may be explained by the challenges in evaluating complex interventions.[44-46]

CHALLENGES TO THE ECONOMIC EVALUATION OF INTEGRATED CARE INTERVENTIONS

There are two main reasons why standard cost-effectiveness analysis may be less appropriate for the evaluation of integrated care: 1) the complexity of the interventions, and 2) the focus on improving outcomes across the triple aim. Firstly, integrated care interventions are complex interventions as they are, by definition, multi-faceted. Complex interventions possess several of the following characteristics: targeting not only individuals with care needs but also groups of professionals providing care or involved in the management of the care process, targeting stakeholders at different organisational levels, consisting of various interacting components, having a variety of intended outcomes, and continuously changing over time to better tailor the intervention to the needs and improve the outcomes, often in learning feedback loops. Compared to single interventions like a drug treatment, the effectiveness of complex interventions is impacted to a greater extent by the behaviour of those delivering and those receiving the intervention.[47,48] To evaluate such complex interventions, it is crucial to fully comprehend what the intervention consists of, how and when it works, and how it interacts with the context in which it is implemented.[48-50]

Secondly, as integrated care interventions aim to affect a wide array of outcomes, current economic evaluations may not fully capture the effectiveness of integrated care.[51] In many countries including the Netherlands, measuring Quality-Adjusted Life Years (QALYs) has become the standard to inform decision-making about reimbursement of innovative interventions, as it has the advantage of offering a common scale that can be used across different clinical areas and treatment options.[52,53] The QALY is a generic measure of disease burden which combines both survival and health-related quality of life into a single index.[52] To compare multiple interventions, the Incremental Cost-Effectiveness Ratios (ICERs) can be put side by side.[54] The ICER is the ratio of the difference in costs between the intervention under evaluation and the next best alternative (often usual care) and the difference in effects between those two, i.e.:

$$ICER = \frac{C_i - C_u}{E_i - E_u}$$

where C_i represents the costs related to the intervention, C_u the costs related to usual care, E_i the effects of the intervention, and E_u the effects of usual care.

The ICER is an indicator of efficiency in and of itself, but it can also be compared to a threshold value that represents the maximum acceptable ICER. When the ICER is below the threshold, the intervention under evaluation is more efficient in generating health than the interventions that would be displaced if the new intervention was introduced in the healthcare system. When the ICER is above the threshold, the intervention is less efficient.[52]

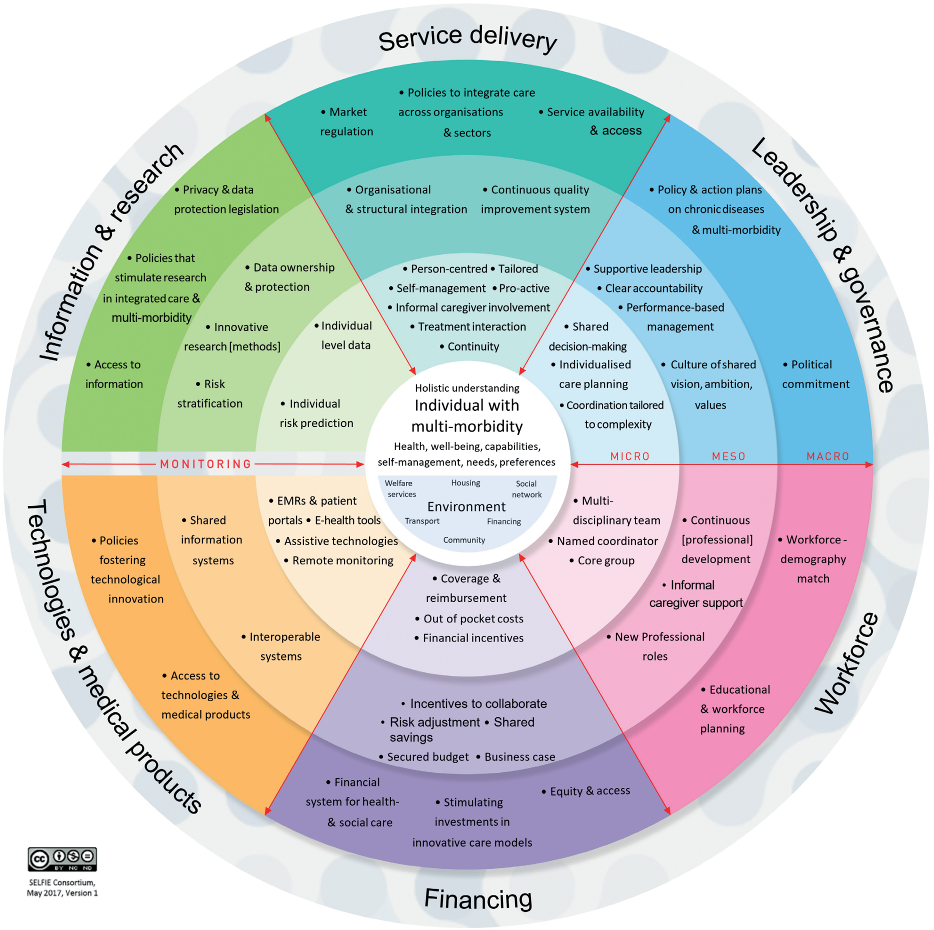
One of the main objections to standard economic evaluations of integrated care for the frail elderly, is the sole focus on health-related quality of life and survival outcomes, combined into the QALY. There are two main reasons why this is not sufficient to measure the impact of integrated care in frail elderly, one is related to the target population and the other is related to the aim of such interventions. Regarding the first, there is evidence that QALYs are not sufficient to measure what is truly important for frail older persons.[42] Outcome measures that underpin health-related quality of life may not be sensitive to improvements in the lives of frail elderly as these generally measure quality of life in terms of health status, focusing on mobility, ability to self-care, ability to perform usual activities, like in the EQ-5D.[55] However, improvements in health status may not be feasible in frail elderly and other outcomes than physical health may become more important for life satisfaction.[56] This includes outcomes beyond health, such as maintaining autonomy, enjoyment of life, resilience, and social relationships. Regarding the second reason, integrated care interventions explicitly aim to improve three categories of outcomes, i.e., the Triple Aim, including population health, patient's experience with the care process, and costs/efficiency.[57] Hence, integrated care interventions may be effective according to persons participating in the intervention or according to the predefined goals, but not according to current cost-effectiveness standards. Therefore, it is essential to broaden the scope of outcome measurement from health status to well-being and include process-related outcomes as well. The growing recognition that the scope of outcome measurement needs to be expanded is also illustrated by the development of other preference-based outcome measures that focus on social care or well-being, such as the Adult Social Care Outcomes Toolkit (ASCOT), ICEpop Capability Measure (ICECAP) for Older persons and for Adults and the novel EQ-Health and Well-being.[58-60]

MULTI-CRITERIA DECISION ANALYSIS

In this thesis we investigate how MCDA could be used to provide information on the added value of an integrated care intervention, in a situation where a standard economic evaluation was expected to be insufficient to capture the relevant benefits of the intervention. MCDA was defined as *"a set of methods and approaches to aid decision-making, where decisions are based on more than one criterion, which make explicit the impact on the decision of all the criteria applied and the relative importance attached to them"*.[61] Thereby, MCDA intends to enhance transparency, consistency, accountability, credibility, and acceptability of decision-making by assessing alternatives based on explicit aims and outcomes that are identified and deemed important for the decision problem at hand.[62,63] The use of MCDA in healthcare is relatively new but the interest in MCDA to inform decisions regarding healthcare interventions is increasing.[62] MCDA provides two additional sources of information compared to economic evaluations based on QALYs. Firstly, it can include outcomes beyond quality of life and longevity. This is especially relevant for interventions that do not lead to gains in life years but to gains in well-being that cannot be measured adequately by a questionnaire that adopts a definition of quality of life that primarily focuses on health status. It is also relevant when decision criteria other than improvement in health and well-being are important. In MCDA, a more holistic understanding of value can be adopted, which better captures what integrated care aims to achieve: improved health and well-being, experience with care, and efficiency. Secondly, the outcomes included in the MCDA are weighted according to different stakeholder groups to determine their relative importance. In value-based methods of MCDA, all relevant outcomes, including conflicting ones, can be aggregated into an overall value score by combining the performance of an intervention in terms of outcomes with importance-weights for these outcomes. In this thesis, the Multi-Attribute Value-based Theory (MAVT) of MCDA was adopted because of the comprehensiveness and robustness of this method.[64] This means that we apply a multi-attribute value function to obtain an overall value for the intervention and its comparator.

To adequately conduct an MCDA, seven steps are commonly distinguished, which were also followed in this thesis.[65] In Step 1 the decision context needs to be established to better understand the intervention and the context in which the intervention takes place. This is important as integrated care programmes are complex multi-faceted interventions that are often viewed as a black box.[66] When the content of the box is unknown, it hampers the understanding of why an intervention succeeds or fails. To guide the comprehensive description of an integrated care intervention, the conceptual framework for integrated care for persons with multi-morbidity that was developed in the Sustainable Integrated Care Models for Multi-Morbidity: Delivery, Financing and Performance (SELFIE) project can be followed, see **Figure 1**. It consists of six components that should be addressed at the micro-, meso-, and macro level: 1) Service delivery, 2) Leadership and Governance, 3) Workforce, 4) Financing, 5) Technologies and Medical products, and 6) Information and Research.[66] The core of the framework puts the individual with multi-morbidity and his/her environment central and forms the basis for a holistic understanding of the individuals' health and well-being, capabilities, self-management abilities, needs, preferences.

• **Figure 1.** SELFIE conceptual framework for integrated care for persons with multi-morbidity



The decision-context also includes the setting in which the intervention is implemented, the stakeholders involved, and the decisions that needs to be made. In this step the aims of the MCDA should be addressed and the alternatives (i.e., the intervention and its comparator(s)) need be determined. Moreover, the decision context refers to whether the results of an MCDA lead to decisions regarding the scaling-up of an intervention, continuation, or long-term reimbursement (i.e., its financial sustainability).

In Step 2, relevant outcome measures need to be identified and structured. These outcome measures will be used to measure the performance of an intervention and should therefore align with what the stakeholders, whose value judgements are considered relevant, view as most important. Furthermore, to be eligible to be included in an MCDA the outcome measures need to meet the following conditions:

1. *Relevance*, i.e., outcome measures should be relevant to persons with multi-morbidity and other stakeholders;
2. *Completeness*, i.e., all important outcome measures need to be included;
3. *Non-redundancy*, i.e., no overlap between the different outcome measures;
4. *Operationalizability*, i.e., is it feasible to measure how well an alternative performs on each outcome measure;
5. *Preferential independence*, i.e., the weight on one outcome should be independent from the performance score on another outcome measure;
6. *Sensitivity to intervention-effects* within the timeframe the evaluation takes place in.

We opted to use mainly patient-reported outcome and experience measures (PROMs and PREMs), and we preferred to use original and validated instruments above self-constructed scales. The core set of outcome measures is structured according to the goals of the Triple Aim.

In Step 3 the performance of an intervention and its comparator needs to be assessed by means of (empirical) evaluation. The performance is measured on the core set of outcome measures identified in the previous step. Measuring performances comes with the challenge of setting up an appropriate study design that allows for causal inferences, as randomisation is often not possible. Therefore, in this study a quasi-experimental study design was chosen, comparing frail elderly of GPs who provide the integrated care intervention to frail elderly of GPs who do not provide the intervention. To account for any differences between the groups at baseline, propensity score matching was applied. A propensity score was estimated using logistic regression modelling, which reflects the '*propensity*' (i.e., probability) an individual is assigned to the intervention based on observed baseline characteristics. This propensity score can then be used to either match the intervention and control group, or to weight the sample using inverse probability weighting.

Step 4 focuses on eliciting weights for the outcomes that represents their relative importance according to the most relevant stakeholders. In this thesis weights were elicited using discrete choice experiments (DCE), for five stakeholder groups: patients, informal caregivers, professionals, payers, and policy makers. In the DCE, respondents were presented with 18 choice tasks that each consisted of two hypothetical care programmes and asked which care programme they prefer. The description of the care programmes systematically differed in terms of performance which forced respondents to trade-off these outcome measures. Collecting weights for different stakeholder groups allows for comparisons between different perspectives and thereby for more nuanced and transparent statements about the effectiveness of an intervention.

Step 5 of performing an MCDA encompasses bringing together the performance scores (derived in step 3) and the relative importance weights (derived in step 4) to create an overall value score for both the intervention and the comparator, for example by applying the 'weighted sum approach'. This means that each performance score is multiplied with its respective weight, and subsequently, all weighted performance scores are summed. A stylistic example of an MCDA table, including weight sets for two stakeholder groups, is given in **Table 1**. The overall value scores can be compared directly and the intervention with the highest value score is preferred. The preferred intervention can differ between stakeholder groups based on differences in their weight sets.

In Step 6, sensitivity analyses should be undertaken to address the uncertainty in the MCDA value scores. To adequately include the joint uncertainty around the performance scores and the relative importance weights, we conducted probabilistic sensitivity analysis (PSA) using Monte-Carlo simulation.[68] Results can be presented in an innovative curve, which we called a Conditional Multi-Attribute Acceptability Curve (CMAC). The CMAC is inspired by the Cost-Effectiveness Acceptability Curve (CEAC) as being used in cost-effectiveness analysis. It shows the probability that an intervention is the preferred alternative (i.e., has a higher overall value score) while the budget impact remains below a set threshold. This threshold represents the total budget that is available for allocation to either the intervention or the comparator, for the treatment of a target population of a given size.

In the final step of MCDA, Step 7, results are deliberated upon, which is an integral part of performing an MCDA and showcases the main advantages of MCDA. That is, MCDA provides the possibility to rationalise the process of priority setting and explicitly discuss with stakeholders what drives their priorities. The discussion includes, among other things, the specification of the decision to be made, the choice of outcome measures, the performance on said outcome measures, and the relative importance-weights assigned to the outcomes by different stakeholders. This has the potential to improve transparency, consistency, accountability, credibility, and acceptability of the decision-making process.

- **Table 1.** Simplified example of an MCDA table, including 4 outcome measures and 2 stakeholder perspectives

Core set of outcomes	**Weights[1]**		**Standardised performance[2]**		**Weighted performance scores[3]**			
					Patients		**Professionals**	
	Patients	**Professionals**	**Integrated care**	**Usual care**	**IC**	**UC**	**IC**	**UC**
Health & well-being								
Psychical functioning	0.100	0.200	0.500	0.700	0.050	0.070	0.100	0.140
Enjoyment of life	0.450	0.100	0.800	0.600	0.360	0.270	0.080	0.060
Experience with care								
Person-centeredness	0.300	0.250	0.800	0.600	0.240	0.180	0.200	0.150
Costs								
Total health- and social care costs	0.150	0.450	0.200	0.500	0.030	0.075	0.090	0.225
Overall value score[4]					**0.680**	0.595	0.470	**0.575**

Clarification of terms used in the MCDA table:

[1] *Weights: hypothetical relative importance weights obtained in DCE for, in this example, two stakeholder groups: 'Patients' and 'Professionals'*

[2] *Standardised performance: hypothetical performance scores of an integrated care programme and usual care, as measured in of example an empirical evaluation. Performance scores are measured on the natural scale of the instrument and subsequently standardised using the method of relative standardisation, with the following formula:*

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

where x = performance score on the natural scale, a = integrated care, b = usual care, j = outcome j

[3] *Weighted performance scores: aggregation of standardised performance scores using weights for each stakeholder group*

[4] *Overall value score: sum of all weighted performance scores*

Interpretation of MCDA table:

Results show that according to patients integrated care is preferred. This can be attributed to the higher performance scores for enjoyment of life and the experience with care outcome person-centeredness, and the corresponding weights. Professionals preferred usual care, which is mainly caused by the higher performance scores for physical functioning and costs, and the high relative importance weight for costs.

RESEARCH SETTING

The research for this thesis was part of the European Horizon2020 Project "Sustainable Integrated Care Models for Multi-Morbidity: Delivery, Financing and Performance" (SELFIE), see **Box 1**.

Box 1

SELFIE was a Horizon2020 funded EU project that aimed to contribute to the improvement of person-centred care for persons with multi-morbidity by proposing evidence-based, economically sustainable, integrated care programmes that stimulate cooperation across health and social care and are supported by appropriate financing and payment schemes. More specifically, SELFIE aimed to:

- Develop a taxonomy of promising integrated care programmes for persons with multi-morbidity;
- Provide evidence-based advice on matching financing/payment schemes with adequate incentives to implement integrated care;
- Provide empirical evidence of the impact of promising integrated care on a wide range of outcomes using Multi-Criteria Decision Analysis;
- Develop implementation and change strategies tailored to different care settings and contexts in Europe, especially Central and Eastern Europe.

Seventeen promising integrated care programmes for persons with multi-morbidity are being evaluated in SELFIE using MCDA and a common set of core outcomes as well as programme-type specific outcomes. The latter depend on whether a programme is i) a population health management programme, ii) a programme targeting frail elderly, iii) a programme targeting persons with problems in multiple life domains, or iv) an oncology or palliative care programme.

The SELFIE consortium included eight organisations in the following countries: the Netherlands (coordinator) (NL), Austria (AT), Croatia (HR), Germany (DE), Hungary (HU), Norway (NO), Spain (ES), and the United Kingdom (UK).
(www.SELFIE2020.eu) [Grant Agreement No 634288]

For this thesis, the Care Chain Frail Elderly (CCFE) programme, as implemented in southeast Brabant, serves as a case study to demonstrate the seven steps of MCDA. The CCFE is an integrated care programme targeted at the frailest of elderly still living at home that receive care from their general practitioner (GP). The programme combines common elements of integrated care, such as performing a holistic assessment, individualised care planning, multidisciplinary care, care coordination, and/or case management, with innovative components, such as inviting the frail elderly and their informal caregiver to participate in the multidisciplinary team meeting and funding the programme through a new bundled payment model. The overall goal of the CCFE is to provide person-centred integrated care, coordination, and case management to support frail elderly in living at home for as long as possible. Additionally, from the payers' perspective, the programme should decrease the demand for secondary care, postpone nursing home admissions, and reduce health care costs for persons in this stage of life.

AIM AND OBJECTIVES OF THIS THESIS

This thesis aims to advance the economic evaluations of complex interventions in integrated care for persons with multi-morbidity, with a specific application to frail elderly. Therefore, the objectives are as follows:

1. *To unravel the perceptions of persons with multi-morbidity about good health and quality care*
2. *To empirically evaluate a complex integrated care intervention for frail elderly using Multi-Criteria Decision Analysis*

This thesis draws on scientific literature, focus groups, data collected in the SELFIE project, interviews, registry and claims data, and data collected with the newly developed SELFIE questionnaire. The research questions were examined from multiple perspectives, including those of persons with multi-morbidity and health- and social care providers, using a combination of qualitative and quantitative methods. An overview of the methods used to address the thesis' research objectives, and the steps in the MCDA process that the chapters correlate with, is provided in **Table 2**.

- **Table 2.** Methods used to address the research objectives

Objectives	Chapter	Method	Seven steps of MCDA
1. *To unravel the perceptions of persons with multi-morbidity about good health and quality care*	2	Focus groups	Step 2: Identifying outcome measures
	3	Discrete choice experiment	Step 4: Eliciting weights
2. *To empirically evaluate a complex integrated care intervention for frail elderly using Multi-Criteria Decision Analysis*	4	Thick description	Step 1: Describing the decision context
	5	Quasi-experimental study	Step 3: Measuring performance Step 5: Creating an overall value score Step 6: Sensitivity analysis
	6	CEA and MCDA	Step 6: Sensitivity analysis
	7	Deliberation	Step 7: Deliberation

Outline of this thesis

In the first part of this thesis, **Chapter 2 and 3** take a more general approach in focusing on what is (most) important in good health and care for persons with multi-morbidity. **Chapter 2** presents the results from focus groups with persons with multi-morbidity in which the question is addressed how participants define 'good health and well-being' and a 'good care process'. Chapter 2 addresses step 2 of the MCDA process, identifying and structuring a core set of outcomes. **Chapter 3** takes a quantitative approach to assess the relative importance of the core set of outcomes using discrete choice experiments. This relates to step 4 of MCDA: eliciting weights to determine the relative importance of the outcomes.

In the second part of this thesis, the chapters will zoom in on the case study CCFE to empirically evaluate its added value. **Chapter 4** gives insights into the complex integrated care programme CCFE, using a thick description approach that includes document analysis and interviews with multiple stakeholders. This chapter pertains to establishing the decision context (step 1). Next, in **Chapter 5**, the results of the MCDA of the CCFE are presented. It presents the results of a 12-month quasi-experimental study among a large group of frail elderly either receiving the CCFE or usual care. It combines the performance of the CCFE on the core set of outcomes established in Chapter 2, with the weights elicited in Chapter 3, within the elaborately described context of the care programme in Chapter 4 that informed the design of the empirical evaluation. This chapter relates to steps 3, 5 and 6 of performing an MCDA, namely, (3) measuring the performance of the CCFE and its comparator, (5) creating an overall value score, and (6) performing sensitivity analysis. **Chapter 6** then further addresses the joint uncertainty present in MCDA by constructing the CMAC (step 6: sensitivity analysis). Moreover, it explores how MCDA can complement cost-effectiveness analysis by including outcomes beyond health and longevity and the preferences of multiple stakeholders, to aid local-level decision-making in elderly care.

Finally, **Chapter 7** describes the main findings of this thesis, and addresses their implications for future policy, practice, and research in the field empirical evaluations of complex integrated care for persons with multi-morbidity.

ABBREVIATIONS

Abbreviation

ATT	Average Treatment effect on the Treated
CBA	Cost-Benefit Analysis
CCA	Cost-Consequence Analysis
CCFE	Care Chain Frail Elderly
CE-plane	Cost-Effectiveness-plane
CEA	Cost-Effectiveness Analysis
CEAC	Cost-Effectiveness Acceptability Curve
CMAC	Conditional Multi-attribute Acceptability Curve
COREQ	Consolidated criteria for reporting qualitative research
DCE	Discrete Choice Experiment
EQ-5D-5L	EuroQoL - 5 dimensions - 5 levels
GP	General Practitioner
Hr-QoL	Health-related Quality of Life
ICER	Incremental Cost-Effectiveness Ratio
ISPOR	The Professional Society for Health Economics and Outcomes Research
IPW	Inverse Probability Weighting
MCDA	Multi-Criteria Decision Analysis
MDT	Multidisciplinary Team
OECD	Organisation for Economic Co-operation and Development
PREMs	Patient-Reported Experience Measures
PROMs	Patient-Reported Outcome Measures
PSA	Probabilistic Sensitivity Analysis
PSM	Propensity Score Matching
QALY	Quality-Adjusted Life Year
SAB	Stakeholder Advisory Board
SELFIE	**S**ustainable Int**e**grated Care Mode**l**s for Multi-Morbidity: Delivery, **F**inanc**i**ng and P**e**rformance
SRoI	Social Return on Investment
WHO	World Health Organisation

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> “Not everything that can be counted counts and not everything that counts can be counted”
>
> - William Bruce Cameron

PART 1

Unravelling the perceptions of persons with multi-morbidity about good health and quality care

Chapter 2

Defining Good Health And Care From The Perspective Of Persons With Multimorbidity: Results From A Qualitative Study Of Focus Groups In Eight European Countries

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ABSTRACT

Objectives: The prevalence of multi-morbidity is increasing in many Western countries. Persons with multi-morbidity often experience a lack of alignment in the care that multiple health- and social care organisations provide. As a response, integrated care programmes are appearing. It is a challenge to evaluate such programmes and to choose appropriate outcome measures. To gain insight into meaningful outcomes, focus groups were held with persons with multi-morbidity in eight European countries, in which participants defined 'good health and well-being' and a 'good care process'.

Methods: In 2016, eight focus groups were organised with persons with multi-morbidity in: Austria, Croatia, Germany, Hungary, the Netherlands, Norway, Spain, and the UK (total N=58). Each focus group followed the same two-part procedure: 1) defining a) good health and well-being, b) a good care process and 2) group discussion on prioritising the most important concepts derived from part one and from a list extracted from the literature. Inductive and deductive analyses were done.

Results: Overall, the participants in all focus groups concentrated more on the care process than on health. Persons with multi-morbidity defined good health as being able to conduct and plan normal daily activities, having meaningful social relationships, and accepting and coping with the current situation. Absence of shame, fear, and/or stigma, being able to enjoy life, and overall psychological well-being were also important facets of good health. Being approached holistically by care providers was said to be vital to a good care process. Continuity of care and trusting professionals were also described as important. Across countries little variation in health definitions were found but variation in defining a good care process was seen.

Conclusion: A variety of health outcomes that entail well-being, social, and psychological facets and especially experience with care outcomes should be included when evaluating integrated care programmes for persons with multi-morbidity.

INTRODUCTION

With ageing populations in Western societies, there is an increasing prevalence of multi-morbidity, i.e., the co-occurrence of two or more health problems within one person at one time (van Oostrum et al., 2012; Barnett et al., 2012). Persons with multi-morbidity therefore often require care from different types of professionals. These professionals may work in different healthcare sectors (e.g., primary and secondary care) and may also work in social or community care. It is important that persons with multi-morbidity receive well-integrated care, in order to avoid the risk of fragmentation or overlap in the care received and interactions in treatment (Mair & May, 2014).

Integrated care is defined as structured efforts to provide coordinated, pro-active, person-centred, multidisciplinary care by two or more care providers that effectively communicate and collaborate. There are different integrated care programmes for multi-morbidity being implemented across Europe that may offer a solution to the aforementioned risks that this population faces (van der Heide et al., 2015; Struckmann et al., 2016; de Bruin et al., 2012; Hopman et al., 2017). The evidence-base of such programmes is still limited and findings are not yet wholly convincing (de Bruin et al., 2012; Hopman et al., 2017; Struckmann et al., 2016). Increasing this evidence-base is important for the durability, wider implementation, and more sustainable reimbursement/financing of such programmes. However, it can be questioned whether the current (economic) evaluation framework, in which costs per Quality Adjusted Life Years (QALYs) are calculated, provides sufficient insight into the broad range of outcomes that such integrated care programmes aim to improve. Integrated care programmes are complex interventions: they consist of various interacting components, target individuals but also groups and organisations, have a variety of intended outcomes, are amendable to tailoring through adaptation and learning feedback loops, and their effectiveness is impacted by the behaviour of those delivering and receiving the intervention (Craig et al., 2008). Common generic outcomes such as QALYs may not fully capture what these programmes are actually trying to achieve in persons with multi-morbidity. Their aims may go beyond the improvement of life expectancy and health-related quality of life, and include the improvement of well-being, the maintenance of independence, and increasing satisfaction with the care process.

In a time when the scarcity of resources is evident and evidence-based decisions on the spending of these resources are warranted, it is crucial to set up appropriate evaluations that can be used to convince decision-makers. Outcomes in the evaluations of complex care programmes often correspond at the higher level to the so-called ‘triple aim’: improving population health [and well-being], improving the patient’s experience with care, and reducing cost [increase] (Berwick et al., 2008; Whittington et al., 2015). These higher-level outcomes, however, can be interpreted in different ways. Health can be defined as the absence of disease (Boorse, 1977), or a wider perspective can be applied whereby health is seen as the complete physical, mental, and social well-being beyond merely the absence of disease (WHO, 1946). More recent definitions turn health into a more active term, as the ability to adapt (Huber et al., 2011) and as a 'meta-capability' that can be used to attain human value (Venkatapuram, 2011). Similarly, the experience with care can include many different aspects, such as the extent of person-centeredness or to which care is proactive (e.g., two domains of the Patient Assessment of Chronic Illness Care (PACIC) instrument, Glasgow et al., 2005).

Different perspectives and approaches can be taken when operationalising the triple aim and meaningful outcomes of integrated care programmes. In the current study we aim to have persons with multi-morbidity define good health or well-being and a good care process, and identify what they find most important in each; thus focusing on two of the three 'triple aims'. We look at this from a cross-country perspective by conducting focus groups in eight European countries involved in the SELFIE research project on integrated care in multi-morbidity: the Netherlands, Austria, Croatia, Germany, Hungary, Norway, Spain, and the UK (see **Box 1**). Using a qualitative focus group approach encourages interaction between persons to take place that may allow for novel concepts and themes to arise.

METHODS

Focus groups were chosen because we were interested in the perspectives of persons with multi-morbidity themselves when it comes to health/well-being and care. The qualitative focus group methodology allows for novel concepts to arise, and interaction between persons can strengthen this process. The Consolidated criteria for reporting qualitative research (COREQ) checklist was used to structure this manuscript (Tong et al., 2007). Eight focus groups were conducted with persons with multi-morbidity, one in each SELFIE partner-country. The Dutch SELFIE team provided all other partners with a protocol on how to conduct and report on the focus groups, held a teleconference with each partner to discuss the protocol, and provided additional support throughout the process. All focus groups were organised between spring and fall of 2016.

Box 1

SELFIE was a Horizon2020 funded EU project that aimed to contribute to the improvement of person-centred care for persons with multi-morbidity by proposing evidence-based, economically sustainable, integrated care programmes that stimulate cooperation across health and social care and are supported by appropriate financing and payment schemes. More specifically, SELFIE aimed to:

- Develop a taxonomy of promising integrated care programmes for persons with multi-morbidity;
- Provide evidence-based advice on matching financing/payment schemes with adequate incentives to implement integrated care;
- Provide empirical evidence of the impact of promising integrated care on a wide range of outcomes using Multi-Criteria Decision Analysis;
- Develop implementation and change strategies tailored to different care settings and contexts in Europe, especially Central and Eastern Europe.

Seventeen promising integrated care programmes for persons with multi-morbidity are being evaluated in SELFIE using MCDA and a common set of core outcomes as well as programme-type specific outcomes. The latter depend on whether a programme is i) a population health management programme, ii) a programme targeting frail elderly, iii) a programme targeting persons with problems in multiple life domains, or iv) an oncology or palliative care programme.

The SELFIE consortium included eight organisations in the following countries: the Netherlands (coordinator) (NL), Austria (AT), Croatia (HR), Germany (DE), Hungary (HU), Norway (NO), Spain (ES), and the United Kingdom (UK). (www.SELFIE2020.eu) [Grant Agreement No 634288]

Recruitment

The goal was to recruit 6-8 persons for each focus group, with a mix of gender, age distribution, and types of multi-morbidity (≥ 2 health and/or social problems) being desired. Participants were recruited via patient organisations (AT, DE, HR, NL, NO), medical professional organisations (HR), self-help groups (AT, DE), medical centres (AT, ES, HU), non-profit care organisations (e.g., Red Cross) (AT), and patient and public involvement groups (UK). We specified that we were searching for persons with multiple health- and/or social problems. Participants were also recruited via SELFIE national Stakeholder Advisory Boards (SAB) (AT, DE, HU, NL, NO). Each country has a SAB that reflects on SELFIE findings and consists of five stakeholder groups, ‘5Ps’: Patients (persons with multi-morbidity), Partners (informal caregivers), Professionals, Payers, and Policy makers. At the time that the focus groups were being organised, a SAB meeting had just taken place in each country, with two to four persons with multi-morbidity present. These persons were reached out to for the focus groups and we tried to snowball via their networks.

Participants were reimbursed for their travel costs and in some countries a gratification was made available either as a token of appreciation or to aid recruitment (AT, DE, NL).

Procedure

An extensive protocol was developed (see Supplementary file Box S.1) that was followed in each focus group. The first focus group was held by the Dutch team, who added 'lessons learned' to the protocol to aid the subsequent focus groups. Each country made a protocol in their own language to use during the actual focus group. The focus groups were all held in a meeting room in an office or clinical setting, with two to four researchers present, one being the chair and at least one taking extensive notes throughout the meeting. At least one of these researchers is a co-author on this publication (FL, VS, MK, SCS, AZ, RE, CV, MiH, MB, MRM). The focus groups were also recorded. These researchers had a minimum of a bachelor's degree and experience/training in patient-contact and/or qualitative research.

The focus groups consisted of an introduction and two main parts: 1) defining a) good health and well-being, and b) a good care process, and 2) discussion on most important concepts and creating 'top-10' lists.

At the onset, the researchers welcomed everyone and introduced themselves, as only some researchers present had already been in touch with participants via telephone or email beforehand. The researchers stated their names, current position, and background expertise/occupation. Following this, in the introduction the researchers stated the purpose of the focus group: to discuss what is important from the perspective of an individual with multiple health- and/or social problems when it comes to health and care. The agenda of the focus group (introduction, part 1, part 2) was described. Next, the 'rules' of the focus group were introduced (see Supplementary File Box S.1) (e.g., respectful interaction, phones off). Participants were asked if they had any questions regarding the informed consent, which they had received beforehand, and had time to read this through if they had not done so already, and signed these. The recorder was then turned on and participants were asked to introduce themselves and briefly describe their multi-morbidities.

In part one of the focus group, participants were asked to define good health/well-being and a good care process. First, a discussion about good health/well-being was held, followed by a discussion about a good care process. We started by asking participants to complete the sentence "For me, being in great health means…" and "I'd be really satisfied with all of the care / the overall care that I receive, if…" Answers were discussed and written on flip-over boards. During this discussion a researcher asked triggering questions when needed and tried to focus the discussion on either health/well-being or care. After this, a researcher went through the statements on the flip-over board and the group tried to move from specific examples to general outcomes. These more general outcomes were highlighted / marked on the flip-overs and written on cards. Again a researcher needed to ask thought provoking questions: What do these original statements really boil down to? How can you expand this so that it counts for everyone in the room? Throughout both steps a researcher also could pose the question as to how this is especially relevant or different for multi-morbidity.

After part one, there was a 15-30 minute break. During the break, the researchers arranged the cards with concepts, or outcomes, mentioned during part one (e.g., on tables or white/magnet boards). They also sorted through the a priori made cards from the literature and added those *not* mentioned during part one to the display. These cards were made a priori by the Dutch team, and distributed to each organiser. The outcomes on the a priori made cards stemmed from a large scientific literature review that had been conducted in the SELFIE project (Struckmann et al., 2017). All outcomes included in those publications were ordered according to the triple aim and, where possible, overlap was removed. This resulted in a list of 77 potentially relevant outcomes (51 health/well-being, 22 experience, 4 cost) (see Supplementary File Box S.2). Each focus group organiser had been asked to translate these outcomes to their respective languages and write them on cards, using different colours for each triple aim.

In part two, the researchers briefly explained to the group that the cards from part one were now on display, and that they also added novel cards on the basis of findings in the literature. These new concepts were mentioned one-by-one and, where unclear, explained. Participants were asked to look at all the concepts. They were asked to write down the ten concepts that were most important to them on a sheet of paper. Hereafter, a discussion was opened as to what was on everyone's 'top-10' list.

Analyses

All focus groups were recorded and extensive notes were made, pictures of flip-overs and cards were made, and the top-10 lists were collected. In two cases, verbatim transcriptions were also made (HR, NO). Reports were made of each focus group following a predefined template: structure of the focus group, recruitment, participant characteristics, reflection on the process, findings part 1, findings part 2, and conclusion/discussion.

Three analytical steps can be distinguished. First, throughout the reporting and summarising done for each focus group, the co-authors analysed and reflected on their respective focus group. This analysis was thus done by the researchers present at the focus group. Next, these reports were analysed independently by the first, second, and second-to-last author from the Dutch team. During this analysis step themes and priorities, corresponding to part one and two, were extracted from each focus group and these were compared across focus groups. Third, these themes and priorities were discussed amongst all co-authors, thus including the authors of the initial reports that were present at the focus groups. The analysis of part one was done applying an inductive approach, part two was done both inductively and deductively as concepts from the literature had been used here and concepts/outcomes were clustered and categorised. No specific qualitative analysis software was used in analysing the data.

The goal from the outset was to conduct one focus group per country, thus data saturation was not discussed a priori. As in most cases no verbatim transcripts were made, these were not returned to participants. However, in some cases, participants were emailed after the meeting to thank them for their participation (DE, ES, HU, NL, NO, UK) and also in some cases were sent notes (NL). About one year after the focus groups, all participants were sent an update on the overall focus group results across countries.

Ethics statement

All participants signed an informed consent form. This form was developed by the Dutch team on the basis of the WHO informed consent for qualitative research (WHO). This was translated by each country into their respective languages. The informed consent consisted of information on the study, the purpose of the research, type of research, participant selection, voluntary participation, procedure, duration, potential risks, benefits, reimbursements, confidentiality, sharing of the results, right to refuse / withdraw, and contact information.

RESULTS

Participant and focus group characteristics

In total 58 persons participated across the eight focus groups. Only in some cases specific persons not participate, this was because we could not get in touch with these persons, they had a conflicting agenda/schedule (e.g., holiday), were too busy, or in some cases had to cancel last minute due to illness or last minute (health-care) appointments.

In all focus groups six or seven persons participated, except in Germany where 12 persons participated. During part two of the focus group in Germany, two subgroups were created to ease discussion. The mean age per focus group is presented in Table 1 below. The overall mean age was 65 years (range 31-86).

During the focus groups most frequently 3 researchers (min 2, max 4) were present, whereby 1 led [part of] the discussion and 1 took notes (total researchers present = 20, M/F = 4/16). The focus groups took 2.5 hours on average (min 2, max 3).

The majority of persons named three morbidities during the introduction of the focus groups (**Table 1**). Health problems were categorised according to the WHO 2010 International Classification of Diseases (ICD-10) (WHO, 2010). For an overview of the specific morbidities mentioned by each participant at the start of the focus groups see Table 2. Across countries, diseases of the circulatory system, musculoskeletal system, and endocrine disorders were most common morbidities amongst participants (Table 2). Specific examples of frequently mentioned diseases were high blood pressure, rheumatism, arthritis, and diabetes mellitus. It is of note that only in the UK mental health problems were explicitly named at the start of the focus groups (**Table 2**).

Table 1. Focus group participant characteristics

	# (M/F)	Mean age (Min-Max)	Mean number of morbidities
Austria (AT)	7 (5/2)	72.9 (62-84)	2.3
Croatia (HR)	7 (4/3)	51.7 (31-69)	3
Germany (DE)	12 (4/8)	62.4 (37-78)	3.9
Hungary (HU)	6 (1/5)	64.5 (47-78)	3
Netherlands (NL)	7 (5/2)	66.3 (53-75)	5
Norway (NO)	7 (2/5)	65.4 (42-76)	3.1
Spain (ES)	6 (5/1)	70.2 (60-81)	2.7
United Kingdom (UK)	6 (4/2)	68.8 (58-86)	4
Total	58 (30/28)	65.3 (37-86)	3.4

Table 2. Morbidities of participants per country categorised according to the ICD-10

	Total	Austria	Croatia	Germany	Hungary	Netherlands	Norway	Spain	UK
Neoplasms	8	3	1	2		1		1	
Diseases of the **blood** and disorders involving the immune mechanism	3		2				1		
Endocrine, nutritional, and metabolic disease	31	2	1	6	5	5	4	3	5
Mental and **behavioural** disorders	6								6
Diseases of the **nervous** system	20	1	2	12	2		1		2
Diseases of the **eye** and adnexa	7				1	6			
Diseases of the **ear** and mastoid process	5			3			1		1
Diseases of the **circulatory** system	44	1	5	12	8	7	4	4	3
Diseases of the **respiratory** system	17		1			6	4	6	
Diseases of the **digestive** system	10	1	1	1	1	2	1	1	2
Diseases of the **skin** and subcutaneous tissue	3	1	1						1
Diseases of the **musculoskeletal system** and connective tissue	36	6	6	9	1	4	5	1	4
Diseases of the **genitourinary** system	4					4			
Congenital malformations, chromosomal abnormalities	1	1							
Symptoms, signs, **abnormal clinical and lab findings**, **not else classified**	2			2					
Injury, **poisoning** and other consequences of external causes	1						1		

Note: ICD-10: International Statistical Classification of Diseases and Related Health Problems, 10th revision.

Defining health and care (part 1)

Defining health

Across all focus groups the idea of good health being the ability to do **'normal' daily activities** was mentioned. This for example included activities such as going outside and undertaking activities (ES), being able to use a computer (UK), and doing physical activities within realistic reach:

> *"…Also being active is necessary for well-being. I was always very athletic nowadays only limited, but it works if you adapt your activities to your physical ability." (P2; AT)*
>
> Response: *"That's true, though I cannot climb mountains anymore, but I can walk through different parks and I can still use the stairways." (P7; AT)*

In three focus groups it was also said that especially in the case of multi-morbidity it is not only about doing such daily activities, but being able **to plan** them and **structure** them yourself (DE, NO, UK). This is related to having realistic expectations about what one can do and relates to acceptance. **Acceptance** was mentioned in terms of self-acceptance and acceptance by others (AT, DE, HU, NL). For example:

> *"Part of it is acceptance, I can do what I can do, and I should leave the other things." (P4; NL)*
>
> *"How to handle the pain and the disease plays an important role. Integration of your diseases in your daily activities, accepting the pain and especially not feeling bad all the time although the diseases are permanent is a big step." (P11; DE)*
>
> *"For me, I even feel good, if no additional things come. In my case, all of my conditions and problems that I was dealing with are considered as end-stage or final stage, and this is accepted. Therefore if my condition is not worsening, then I am fine." (P5, HU)*

Daily activities are also linked to the desire to maintain **social relationships** and **participation** in society (AT, DE, ES, HR, HU, NL, NO).

> *"I want to be consciously active in society. With everything, doing what I want to do, being useful for others." (P7; NL)*
>
> *"For me, good health means being psychosocially active…" (P1; HR) (i.e., having social relationships that are meaningful, being an active participant in everyday life)*

In many countries good health was also associated with the **absence of shame, discrimination, fear, and/or stigma** (ES, HR, HU, NL, NO, UK). These feelings could relate to the wider public, the patient-provider, or the personal realm. For example, in Spain the examples of fear of walking alone and being vulnerable or shame of being seen with medical apparatuses such as an oxygen machine were mentioned. In Hungary shame was mentioned in the patient-provider relation, e.g., not being able to keep to a diet. In Croatia these concepts were summarised as personal vulnerability. Fear of the future and not knowing how the disease trajectory will go was also discussed (HU, NL, NO). Doubts and worries about sharing updates on ones' diagnoses with family and the impact and burden that might have was mentioned (AT). In the Spanish focus group the discussion on fear and shame was heated, as persons dealt and coped with this issue differently. This relates to a point that also came up, of not only accepting the health problems, but also **coping**, being resilient, managing, and having responsibility for the diseases themselves, which may be perceived to be especially difficult in the case of multi-morbidity (DE, HU, NL, NO, UK).

> *"Oh yeah, I think if you have multiple [diseases] it just adds to the whole workload really, how to cope with different things, oh yeah." (P1; UK)*

The general idea of good health being defined as **feeling safe** also was discussed in various focus groups (ES, HU, NL, NO, UK). This can be seen as the result of being absent from the aforementioned negative states (e.g., fear). Feeling safe was also discussed in terms of trusting professionals (discussed more below). In Norway, feeling safe also extended to the economic realm:

> *"...fear, fear for one day having to give up your work for example ... if I don't manage any more, if I am unable to work anymore, then we do not have the economic resources to live here anymore, this was in my thoughts when the illness hits me and then fear..." (P2; NO).*

Lastly, during the open discussion in part one, participants mentioned having a positive frame of mind, being able to enjoy life, and the importance of **psychological well-being** was mentioned in all focus groups.

> *"I think psychological problems ought to be mentioned. Many people with chronic illnesses are also struggling psychologically. It could be because you have bad conscience, because you are dreading something, because you do not know if you will manage and suffer from performance anxiety". (P1; NO)*

> *"I want to enjoy my life, even though I've these diseases. That means, just being full of life." (P4; AT)*

Defining care

When considering the wide array of themes discussed when defining health/well-being, it is not surprising that good care was defined by many persons as being approached as a whole person and **being treated holistically** (AT, NO, UK).

> *"I wish that people treat me in a respectful manner, because it's true, I am sick, but the disease is not me. I don't want to be reduced to my diseases." (P2; AT)*

> *"So I would have wished for a doctor that, to put it this way, had the overview of the whole human being, that he shouldn't treat a heart disease just in isolation, you have another disease, and a third..." (P2; NO)*

Persons explained that being approached holistically also means receiving **holistic support**, including informal caregiver support (HR, NL, NO, UK), good information provision, and especially **emotional and psychological support.** Participants mentioned that support should take the form of more extensive, easily accessible, and timely information on the health problems at hand and medication, but also psychological support, support with self-management, and self-help groups for example (AT, DE, ES, HR, HU, NL). Concerning emotional and psychological support specifically, this is needed even when this is not the 'main' problem at hand (AT, ES, HR, NL, UK). For example, in the Netherlands participants mentioned that in the case of multi-morbidity it takes time to accept the new health problems that arise on top of existing health problems and that there should be support for this adjustment period. In the UK a disconnection between the patient- and provider's priorities was seen, as it was mentioned that the healthcare professionals does do not focus on the mental health issues, when they should:

> *"Yeah, for me I'd like the mental health to be bigger...for me all my healthcare professionals see the [physical disease] as the big thing with me, I don't, I see my depression as the big thing because that's what affects me day to day." (P4; UK)*

> *"What I see is that there's no psychological aid. When you are told you are a chronic patient and you have to take a drug all your life, and that this is for all your life, some people are depressed, needs psychological aid..." (P1; ES)*

A wider theme discussed was **trust in professionals and the system**. This is in part linked to the emotional and psychological support, whereby persons felt that two-way trust is needed in the relationship between the patient and provider so that psychological issues can be discussed, the provider really listens, and so that they can embark on the care process together (AT, NL, NO, UK). Related to this, respectful interaction between provider and patient was also often mentioned (AT, DE, NL, NO, UK, HU). Specifically in the UK, the direction of trust was mentioned with participants saying that they needed to be 'believed' by the professionals (P2, P4, UK).

Trust also pertained to being able to rely on the provider in being able to help, based on their skills and knowledge (DE, ES, HR, NL, NO, UK). Participants spoke of having a 'prepared' care provider to talk to, and trusting their expertise and education.

> *"Yeah, but there are difficulties with medication when you've got multiple things…" (P5; UK)*
>
> Response: *"Oh yeah, but you'd expect your GPs to keep on top of that, it is debatable as to whether that actually happens or not." (P1; UK)*

In Hungary, Germany, and the Netherlands the trust also pertained to the professionals in general:

> *"Trust in the medical world, well I'm sceptical about that, a lot of things happen around you, that you think, did they not see that?… Yeah, then you lose faith/trust because of the things that happen to you…" (P2; NL)*
>
> *"If I visit my doctor, I wish I could reproduce what he is doing there, which services are provided and I would like to sign for them. I want to be more informed about what is done. This way, I often cannot trust my physicians." (P8; DE)*

As may be especially relevant in multi-morbidity, issues surrounding **continuity of care** were mentioned in all focus groups. This pertained to clear responsibilities, a clear contact point, transfer and 'after care', good communication, and good collaboration and teamwork. These points about continuity often also related to **sharing information or medical records** between providers and organisations (DE, ES, HR, HU, NO, NL).

> *"The problem is the coordination between the primary health centres with the reference hospitals. The doctor at the hospital should see all the information, and the family doctor as well…" (P5; ES)*

> *"I would expect that all the institutions and all the GPs in this city would be connected to the same electronical [IT] system. In this case I would not need to go with all those stacks of papers when I see a doctor. (...) There should be a system which can be seen by everyone and not only by some particular segments of the care." (P1, HU)*

Cross-country comparisons in defining good health and care (part 1)

Little variation was seen between countries when defining health (part 1). Themes surrounding the definition of a good care process differed more between countries. Although aspects of access and availability were mentioned in all countries, this concept took a different form between the countries. In some cases this pertained to timely care in terms of waiting lists and time (ES, HU, NL). In Hungary, however, this specifically had to do with the lack of information on the waiting times. Patients emphasised that waiting times were more acceptable if patients knew in advance how long they had to wait.

> *"The worst thing is waiting... They could calculate an order with some gap in between. There are problems with the information sharing" (P6; HU)*

Access in the form of time also referred to professionals having enough time for persons with multi-morbidity in Germany, Hungary, and Norway. Availability also had different definitions per country – in Croatia, Germany, and the UK this was treatment, care, and provider availability and the freedom to choose between them. In Norway, Spain, and the UK this was also geographical- availability and access.

Several themes were only brought up in part one by certain countries. Namely, bureaucracy or the reduction of the burden thereof was mentioned in Germany and the Netherlands, for example with regard to care services that fall within our outside of the insurance package.

> *"There are persons who are physically not able to walk to their physiotherapist anymore. In fact they are in need of transport, but you have to apply for transportation for every single therapeutic unit. These are very difficult circumstances due to too much bureaucracy. This is disastrous..." (P9; DE)*

In Austria participants discussed the need for more self-help and support groups. In contrast, in the Netherlands this was mentioned as a positive aspect already present and available. Participants in Austria were aware that these things exist in other countries, and felt it was missing in their own context.

Lastly, physical surroundings in care provision were mentioned in Hungary that were related to cleanliness, enough personal space, and enough seats.

> *"Because if we consider our life or our homes, we do not like if it is dirty and messy. In a good facility there should be cleanliness, order and discipline." (P2; HU)*

> *"At the … [department] patients need to wait in a very narrow corridor with one row of chairs. There are about three times more patients than chairs. (P5; HU*

Furthermore, in Hungary systematically organised operating procedures in care was an important requirement mentioned by multiple patients. This is made apparent through the fact that the care process in hospitals is sometimes unreasonably long, the staff is in rush, patients do not get clear instructions on the next steps of the procedure, there are redundant diagnostic test, and information is not shared appropriately among professionals.

Most important concepts (part 2)

In all focus groups, in part 2, participants were asked to make a top-10 list of most important concepts. These concepts were those identified in part 1 and supplemented with 'missing' concepts from the literature (see Supplementary File Box S.2). These top-10 lists were discussed. There was large variation within focus groups (i.e., countries) as to what concepts were deemed most important and on persons' top-10 lists. In Supplementary File Box S.3 all concepts written on persons' top-10 lists are presented, including those from the literature and 'novel' concepts derived upon during part 1 of the focus groups.

In **Table 3** below an overview of the most frequently mentioned concepts is presented. These are concepts written on the top-10 lists of at least 10 persons (out of the 58 participants in total) across all countries. The health and well-being concepts social relationships, a positive frame of mind or resilience, enjoyment of life, and maintaining independence were most frequently noted by participants on their top-10 lists. A positive frame of mind or resilience, was mentioned by at least one person in each countries' focus group. Many facets pertaining to good interactions between care providers and persons with multi-morbidity were on the top-10 lists of participants, such as good communication, shared decision-making, and respect. Furthermore, individualised care planning and a holistic assessment and understanding of the problems at hand were aspects of the care process also frequently on the top-10 lists. Lastly, pro-active, prevention-oriented, care was found important by many participants. As can be seen, there is great variety in concepts that participants put on their top-10 lists, both between and across countries. Furthermore, some phrases and words are

Table 3. Most frequently listed outcomes on the 'top-10' lists of focus group participants across the eight focus groups

	# / 58	**Austria (AT)**	**Croatia (HR)**	**Germany[1](DE)**	**Hungary (HU)**	**Netherlands (NL)**	**Norway (NO)**	**Spain (ES)**	**United Kingdom (UK)**
Health and well-being									
Energy & fatigue	**12**	4/7			1/6	4/7	2/7		1/6
Feeling safe	**10**		1/7		3/6	3/7		3/6	
Cognitive functioning	**12**	4/7		1/6		2/7	1/7	3/6	1/6
Maintaining independence	**16**		2/7	5/6	1/6	1/7		1/6	1/6
Enjoyment of life	**16**	2/7		4/6	1/6	3/7	3/7		3/6
Positive frame of mind, resilience	**16**	2/7	3/7	2/6	2/6	1/7	2/7	2/6	2/6
Self-esteem	**11**	1/7	1/7	3/6		3/7	2/7		1/6
Social relationships	**17**	3/7		5/6	2/6	4/7		2/6	1/6
Societal participation	**12**	1/7	3/7	3/6		3/7		1/6	1/6
Experience									
Individualised care planning / tailored care	**13**	1/7		4/6	1/6	2/7	4/7		1/6
Holistic assessment/understanding	**11**	1/7	1/7	4/6	2/6	2/7			1/6
Good communication between provider-patient	**14**	1/7	1/7	2/6		2/7	1/7	3/6	4/6
Shared decision-making provider-patient	**13**	2/7		4/6	1/6	1/7	1/7		4/6
Respectful interaction between provider-patient	**12**	2/7	1/7	5/6					4/6
Shared information between providers	**10**	2/7	1/7	1/6	2/6	2/7		1/6	1/6
Team work between providers	**10**		1/7		2/6		2/7	2/6	3/6
Confidence in knowledge and skills in providers	**10**	1/7	2/7	3/6	2/6	1/7		1/6	
Pro-active, prevention-oriented, care	**13**		1/7	1/6	4/6	4/7			3/6

Note: 1 = Group split into 2, so each time # out of 6 persons, each group discussed either health/well-being and costs or care and costs.

quite specific whereas others are broad; participants were free to determine at what conceptual level they wrote their top-10 outcomes.

DISCUSSION

Main findings

Participants defined good health and well-being in terms of being able to conduct 'normal' daily activities, being able to plan and structure these, and having social relationships and participating in society. Acceptance by oneself and by others, and coping with, one's current health situation was deemed an aspect of good health. Absence of shame, discrimination, fear, and/or stigma from the public, care providers and oneself, and on the other end feeling safe and psychological well-being were also facets of good health. Social relationships, resilience, enjoyment of life, and maintaining independence were considered the *most* important aspects of good health across participants from all countries. A good care process was defined as one whereby persons are approached and supported holistically, with specific attention for emotional and psychological support, there is confidence and trust in professionals and the system, continuity of care is guaranteed, and that information is shared and accessible within a reasonable time. Concepts deemed *most* important were good communication, shared decision-making, and respect between care provider and the person with multi-morbidity, as well as individualised care planning and pro-active, prevention-oriented, care. Little cross-country variation in health themes were found, however, in defining care differences did exist, for example in terms of the exact type of access referred to (e.g., geographical, timely).

Interpreting findings

Several themes brought up during the focus groups were explicitly mentioned to be more relevant for persons with multi-morbidity; this was especially the case when defining care. One such concept was enough time: in Norway persons mentioned that it is difficult when their issues are not immediately visible for a care provider, and that they need time to explain the multiple problems at hand. Also confidence in providers' skills are related to their needing to address or at least be aware of multiple problems and often multiple medications, some of which may go beyond their specialist area of expertise. Also the importance of well-coordinated and smooth transitions (i.e., continuity of care) is particularly so for persons with multi-morbidity, who often cross provider-, organisation- and sector boundaries throughout their care trajectories. In the realm of continuity of care, in many focus groups sharing information for example via shared electronic medical records was explicitly mentioned. It is of note that issues surrounding privacy were not mentioned by participants, although EMRs are in most cases not in place yet due to issues at a wider system level.

Aspects especially to do with defining care seemed relatively unique and extra relevant for multi-morbidity. However, there was a large degree of overlap in the concepts mentioned in defining health in the current focus groups and the existing definitions presented in the introduction by the WHO, Huber and Vankatapuram, and similar studies conducted on defining health. Namely, absence of shame, discrimination, fear, and/or stigma, feeling safe, psychological well-being, and social relationships and participation especially overlap with the mental and social well-being aspects of the 1946 WHO definition of health (WHO, 1946). The importance of mental health outcomes has also been recently found in a study amongst a UK general population on what aspects of the EuroQol 5-dimensions (EQ-5D) are currently missing or receive too little attention, i.e., mental health (Shah et al., 2017). This is also in line with a study by Ebrahimi and colleagues (2012) amongst frail elderly where being able to 'master' daily life (e.g., coping and acceptance) and being happy and satisfied with life (e.g., enjoyment of life) were described as characteristics of health.

Other concepts found in the current focus group relate more to the definition by Huber and colleagues of health as the ability to adapt, i.e., acceptance, coping, resilience (Huber et al., 2011). Also overlap was seen with our finding on the ability to conduct normal daily activities and the aforementioned study on frail elderly where the ability to live the routine life persons are accustomed to was identified as an aspect of health (Ebrahimi et al., 2012). Lastly, especially Venkatapuram's definition of health as the capability to attain human value, seems to overlap with the general idea of the ability to participate in normal daily activities and social participation (a means to attain human value) and enjoyment of life as mentioned throughout the current focus groups (Venkatapuram, 2011).

It was noticeable that, across all focus groups, aspects relating to care were already mentioned in defining good health. It appeared that persons could not always clearly distinguish the two. This might be due to the fact that persons more easily can identify these care-related factors as 'changeable'. When exploring which aspects of health/well-being and care were most important in part two of the focus groups, we saw across countries that these overlapped with those mentioned during the first part of the discussions but also that often concepts were included from the literature-cards. As these were introduced and explained after the break, participants reacted positively to them. This indicates conceptual overlap in what is currently being assessed in evaluations, and thus found in the literature a priori, and what participants of our focus groups found important (Struckmann et al., 2017).

Strengths & limitations

Several limitations in the current study should be mentioned. Only in the UK mental health problems were explicitly mentioned by participants when morbidities were named at the start of the focus groups. This, however, did not seem to have any effect on our findings, as attention for psychological health was deemed an important outcome across focus groups. The difference in explicitly naming mental health problems may relate to culturally-related stigma issues. For example, throughout the Dutch focus group it became apparent that depression issues were also present amongst participants. Also in the German focus groups mental health problems, such as depression, were mentioned as 'side effects' of other health problems.

It was not the goal of the current study to attain a representative sample of participants per country. Participants in some countries may, for example, be the more 'involved' patients. As such they are involved in research more often and may be more familiar with the topics discussed in the focus groups than an average patient. However, considering that our findings are largely in line with expectations based on previous studies, we do not feel that their perceptions of what good health and care is, and priorities therein, systematically differ from other patients.

Initially, the goal of the second part of the focus groups was to reach a group 'top-10', in which participants would discuss their individual lists and try to create a group list. However, due to time restrictions this was not possible. Thus the results of part two reflect each individuals' opinion. We opted to present findings in the results section mentioned by ten or more persons across the focus groups, but this is a relatively arbitrary cut-off point, and the full findings are shown in the Supplementary File Box S.3. The total number of cards from which participants could select concepts from for their top-10 list differed in size depending on from which country they are from. The level of specificity in concepts also differs greatly, for example the overall satisfaction with care process is mentioned, as well as the specific facet that a provider has enough time to listen to a patient (see Supplementary File Box S.3). Quantifying such a qualitative process is thus to be done cautiously. In the Supplementary File Box, concepts are clustered into broader concepts, there are of course different definitions and interpretations for terms, and as many concepts are all interrelated a level of subjectivity is introduced here.

We consider it a strength of the current research that the participating countries in SELFIE differ in geographical size, population [density], culture, and welfare and health care systems – providing a wide scope. Furthermore, it is a main strength that persons with multi-morbidity were so actively approached and included in the current study and in the wider SELFIE study, introduced in Box 1. As will be described below, the results of these focus groups form the predominant input for the core set of outcomes to be included in the SELFIE evaluations of 17 integrated care programmes: participants were given a direct voice and greatly valued this. Another main strength of the current study and great addition to the literature is the fact that we asked participants in our focus groups to not only reflect on health, but also on experience with care. In situations whereby health improvements, especially in the traditional physical and clinical health indicators, are infeasible, focusing on their having a satisfactory and even positive experience throughout the care process becomes more important. And, as was seen in the current study, experience with care is often inextricably linked to the definition of health. Although we did not explicitly incorporate costs in the first part of the focus groups, and thus predominantly focus on two of the three 'triple aims', participants did mention several cost concepts, also in prioritising. In the future, it would be interesting to also have an explicit discussion on what role costs play in, especially, experiences with care.

Next steps

When evaluating integrated care programmes for multi-morbidity, a wide array of health- and care outcomes should be included. Few aspects related to physical functioning, a traditionally used outcome, were found on the top-10 lists of importance, i.e., only 'energy and fatigue' was mentioned by more than 10 persons across all focus groups. Instead, more well-being, social, and psychological facets of health are important and should thus be included in evaluations. This finding overlaps with current trends in conducting more comprehensive evaluations. To this end, in SELFIE a core set of outcomes for the evaluation of 17 promising integrated care programmes for multi-morbidity was developed that places a lot of focus on such outcomes. The core set consists of the following outcomes: physical functioning, psychological well-being, social relationships & participation, enjoyment of life, resilience, person-centeredness, continuity of care, and total health- and social care costs. This core set was developed on the basis of four sources, of which the focus groups described in the current article had the most prominent influence. The other three sources were: i) findings from a literature review conducted to develop a framework on integrated care for multi-morbidity and to identify existing programme evaluations (Struckmann et al., 2017), ii) (inter)national stakeholder advisory board discussions in the eight SELFIE partner countries with Patients, Partners, Professionals, Payers, and Policy makers, and iii) the aims of the aforementioned 17 programmes and current indicators included in these programmes. A set of criteria was used to come to this list (see Supplementary File Box S.4), and some concepts mentioned in the focus group were not included in the core set, because these did not meet these criteria. This is the case for the more system- and cultural-wide concepts, such as absence of public stigma, which cannot be used to assess the performance of one specific care programme.

CONCLUSION

A variety of health outcomes that entail well-being, social, and psychological facets should be included when evaluating integrated care programmes for persons with multi-morbidity. Even more imperative is that experience with care outcomes be included in such evaluations that tackle the complexity of multi-morbidity care provision. In conclusion, important next steps include moving towards harmonising evaluation frameworks and the specific indicators used in such evaluations.

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SUPPLEMENTARY FILES

- **Box S.1:** Extended protocol of the focus group

Introduction

- Purpose: to discuss what is important from the perspective of an individual with multiple health- and/or social problems in health/well-being and in care
- Overarching aim: the results of the discussion will be used to improve the care for persons with multiple health- and social problems
- Agenda explanation
  - 'Rules'
  - No right / wrong answers, only differing points of view
  - Not necessary to agree with others, but must listen respectfully as others share their views
  - Talk to one-another, researchers only have role as moderator in guiding the discussion
  - Privacy / anonymity
  - The discussion will be recorded, and notes will be made
  - Turn off phones
- Informed consent
- Introduction of participants to one-another

Part 1: Defining good health/well-being and care

Step i):

- When you think about your health and well-being: try to complete this sentence / thought: "For me, being in great health means..."
- When you think about the care that you receive, have received, or may receive in the future, try to complete this sentence / thought: "I'd be really satisfied with all of the care / the overall care that I receive, if..."

**Write this question on a large white board / flip-over*

  - Provide examples if participants are having trouble understanding what you mean:
    * ...being able to work full-time. (health)
    * ...being able to spend time with family and friends. (health)
    * ...being able to live for many more years. (health)
    * ...not going to the doctor so often. (health)
    * ...my doctor took more time to explain things to me. (care)
    * ...my partner would always be allowed to join me and discuss with the doctors as well. (care)
    * ...my care plan was discussed with me. (care)

  - * ...I had one overall written care plan. (care)
  - * ...I always knew I could contact one person in case of questions. (care)

**The researcher needs to ask triggering and thought-provoking questions here.*

**The discussion should be about health and well-being in general, not about care specifically (yet).*

**Be careful not to provide too many examples and to steer the answers in one direction.*

**The researcher leading the discussion will be writing these thoughts out on a white board / flip-over.*

**The researchers need to allow the participants to reflect on one-another's ideas.*

**Close the discussion with a final question: i.e., have we missed anything?*

Step ii):

- Once it seems as though all participants have said what is important to them, the researcher will go through the points on the board, and with the help of the participants try to extrapolate what these things mean, in order to go from **specific** examples to **general** outcomes. For example:
  - "For me, being in good health means being able to work full-time", does this mean:
    - * Participating in society?
    - * Being able to support one-self financially? → Self-sufficiency
    - * Having social contacts?
    - * Having a daily structure?
  - "For me, being in good health means not going to the doctor so often", does this mean:
  - Having no acute health problems?
  - Having the ability to deal with minor health problems on your own? (self-management)
  - "I'd be really satisfied with all of the care / the overall care that I receive, if my doctor took more time to explain things to me", does this mean:
    - * Good communication?
    - * Shared-decision making?
    - * More/enough time?
  - "I'd be really satisfied with all of the care / the overall care that I receive, if my partner could always be allowed to join me and discuss with the doctors as well.", does this mean:
    - * Understanding your personal situation?
    - * Involvement of the informal caregiver?
    - * Good communication?

**The researcher needs to write key words that come forth on large notecards and place them in the centre of the table for all participants to be able to see.*
**The researcher needs to ask triggering and thought-provoking questions here – what do the points mentioned in step 1 really mean? What does it come down to? What would it mean for the person sitting next to you? How can you expand this so that it 'counts' for everyone in the room? Try to distinguish between individual opinions and actual group consensus.*
**In asking further questions, try to focus on multi-morbidity – how is this specific to you as a person with multiple health- and/or social problems, would this be different if you had a single disease?*
**For each initial statement, multiple cards can be made.*
**Close the discussion with a final question: i.e., have we covered all elements?*

Break
**Place cards from part one onto the table, sort them by 'triple aim'*
**Sort through original 77 cards, and add cards to the table from these (refrain from overlap, choose card from part one in words of participants over literature-cards)*

Part 2: Selecting most important concepts

- Instructions: "In the discussion during part 1 you've come up with points that are important for you when it comes to defining health and care. As you saw, we've been trying to write this into more general terms on these cards."
- In our research team we've also been looking at concepts that are often mentioned in research/literature as being important, we've added these to the table as well."

**Explain the new concepts in case anything is unclear.*

- "We'd like to ask each of you to look at all of these cards and write down for yourself which 10 are most important to you. This can be a mix of health/well-being, care, and cost cards."
- "Now we'd like you all to discuss your top 10 and try to see whether you can reach a group consensus, and make a top 10 of cards at the centre of the table."

**Ask a participant to first put his/hers down or read his/her list first, through discussion see whether some kind of consensus can be reached.*

Box S.2: List of literature-based outcomes used on cards in part two

Health / well-being

- Biomedical outcomes: e.g., blood sugar, blood pressure, lung function, kidney function
- Life expectancy
- Maintaining independence
- Activities of daily living: e.g. eating, bathing
- Independence from medical aids: e.g., wheelchair, oxygen
- Energy and fatigue
- Pain and discomfort
- Disability
- Physical mobility: e.g., being able to walk
- Work ability: being physically and mentally able to work
- Cognitive functioning: e.g., thinking, memory, concentration and learning
- Anxiety and depression
- Stress
- Worrying
- Listless: no spirit, apathetic, indifferent
- Self-esteem: a good feeling of one-self, self-respect
- Respect from others
- Social relationships: staying in contact with family/friends/acquaintances
- Maintaining social status: in terms of 'societal class', 'occupation', how you think others look at you in society.
- Societal participation: e.g., volunteer work, club memberships, community activities
- Social support: emotional, instrumental and financial assistance from the social network
- Loneliness
- Investing in future health and wellbeing
- Self-efficacy: belief in one's capacity to follow a needed or desired course of action
- Positive frame of mind, resilience: ability to maintain a positive perspective regarding the future, despite setbacks or change
- Coping
- Being able to deal with problems, losses, adverse events in an effective manner
- Health literacy: capacity to obtain, process, and understand information about illness and health
- Compliance/adherence to treatment
- Lifestyle: e.g., smoking, nutrition, physical activity, alcohol and drug use

- Feeling safe
- Enjoyment of life
- Maintaining dignity
- Living comfortably
- Caregiver burden

Experience with care
- Holistic assessment
- Shared decision-making
- Tailored care
- Individualised care planning
- Pro-active, prevention-oriented care
- Clear responsibilities and accountability
- Named coordinator
- Team work and collaboration between professionals
- Good communication
- Shared information between professionals and providers
- Continuity of care: pertains to continuity (e.g., follow-up, monitoring, stability, transfer) throughout time and between professionals and organisations)
- Confidence in knowledge and skills in professionals
- Respectful interaction between provider-patient: feeling of being treated with respect and being ‘heard’
- Geographical access: e.g., facilities close to home
- Physical access: e.g., building is wheelchair accessible
- Timely access: e.g., no/short waiting lists
- Satisfaction with care process

Costs
- Service and support coverage: co-payments/deductible and financial access to services
- Travel and parking costs
- Loss of income – patient
- Loss of income - informal caregiver

Box S.3 Most important concepts in health/well-being, care, and costs per focus group participant per country (part 2 of the focus groups)

	# / 58	**Austria**	**Croatia**	**Germany**[1]	**Hungary**	**Netherlands**	**Norway**	**Spain**	**UK**
Health and well-being									
Physical well-being									
Energy & fatigue	**12**	4/7			1/6	4/7	2/7		1/6
Pain & discomfort	**4**	3/7							1/6
Biomedical outcomes	**3**	2/7			1/6				
Disability	**5**		2/7		1/6		1/7		1/6
Progression free health state*	**2**				2/6				
Physical mobility	**5**		1/7	1/6	1/6	1/7	2/7		
Activities of daily living	**7**		1/7	3/6	1/6	1/7			1/6
Life expectancy	**1**					1/7			
To be active*	**2**						2/7		
Feeling safe	**10**		1/7		3/6	3/7		3/6	
Discrimination*	**3**		3/7						
Cognitive functioning	**12**	4/7		1/6		2/7	1/7	3/6	1/6
Independence									
Being able to choose what you do / want to do*	**1**							1/6	
Moving freely around street *	**1**							1/6	
Independence from medical aids	**3**				1/6			1/6	1/6
Maintaining independence	**16**		2/7	5/6	1/6	1/7		1/6	1/6
Living comfortably	**5**	1/7		2/6		1/7			1/6
*Psychological well-being**	**3**			3/6					
Listlessness	**3**	2/7						1/6	
Loneliness	**2**	2/7							
Empathy*	**1**		1/7						
Stress/worrying	**7**				3/6		1/7		3/6
Anxiety & depression	**2**				1/6				1/6
Enjoyment of life	**16**	2/7		4/6	1/6	3/7	3/7		3/6

- **Box S.3** continued

	# / 58	Austria	Croatia	Germany[1]	Hungary	Netherlands	Norway	Spain	UK
Positive frame of mind, resilience	**16**	2/7	3/7	2/6	2/6	1/7	2/7	2/6	2/6
Creativity*	**2**		2/7						
Investing in future	**1**							1/6	
Coping	**5**	1/7					2/7	1/6	1/6
Maintaining dignity	**9**	1/7			4/6			2/6	2/6
Self-esteem	**11**	1/7	1/7	3/6		3/7	2/7		1/6
Acceptance*	**4**			3/6		1/7			
Look out for possibilities*	**1**						1/7		
Hope	**1**						1/7		
Social relationships & participation									
Social relationships	**17**	3/7		5/6	2/6	4/7		2/6	1/6
Societal participation	**12**	1/7	3/7	3/6		3/7		1/6	1/6
Respect from others	**6**					2/7	2/7	1/6	1/6
To be taken seriously*	**3**						3/7		
Social support	**5**	1/7	1/7				2/7	1/6	
Being able to relate to other people without physical limits*	**1**							1/6	
Feeling useful*	**2**							2/6	
Get something off one's chest*	**2**	2/7							
(Maintaining good) social status	**5**	2/7		3/6					
Not being reduced to the disease*	**6**	1/7					4/7		
To be regarded as whole human being*							1/7		
Seeing past the outside/surface of a person*									
Surrounding area takes care of someone's needs*	**1**	1/7							
Treatments compatible with work*	**1**	1/7							
Work ability	**6**		3/7		1/6	2/7			
Social expectations*	**1**		1/7						

- **Box S.3** continued

	# / 58	**Austria**	**Croatia**	**Germany^1**	**Hungary**	**Netherlands**	**Norway**	**Spain**	**UK**
Daily structure*	**4**			4/6					
Being able to plan*	**2**						2/7		
Burden for informal caregivers*	**3**			2/6			1/7		
*Self-management**	**7**			6/6		1/7			
Lifestyle	**3**	2/7							1/6
Compliance/adherence to treatment	**5**	1/7		2/6	1/6				1/6
Self-efficacy	**6**		2/7		1/6			1/6	2/6
Investing in future health & well-being	**4**		1/7	2/6		1/7			
Health literacy	**7**			5/6	1/6	1/7			
Daily quality of life	**1**						1/7		
Experience									
Individualised care planning / tailored care	**13**	1/7		4/6	1/6	2/7	4/7		1/6
Holistic assessment/understanding	**11**	1/7	1/7	4/6	2/6	2/7			1/6
Good interactions providers-patients									
Good communication	**14**	1/7	1/7	2/6		2/7	1/7	3/6	4/6
Encourage patients*	**1**							1/6	
Psychological support*	**2**			1/6		1/7			
Optimist reactions*	**1**							1/6	
Shared decision-making	**13**	2/7		4/6	1/6	1/7	1/7		4/6
Respectful interaction between provider-patient	**12**	2/7	1/7	5/6					4/6
Trust*	**7**			3/6		4/7			
Predictability of care service delivery* (good info)	**1**				1/6				
Flow of information*	**2**						2/7		
Being listened to and believed*	**2**								2/6

- **Box S.3** continued

	# / 58	Austria	Croatia	Germany[1]	Hungary	Netherlands	Norway	Spain	UK
Good collaborations providers									
Shared information between providers*	**10**	2/7	1/7	1/6	2/6	2/7		1/6	1/6
Data transparency*	**3**			3/6					
Team work between providers	**10**		1/7		2/6		2/7	2/6	3/6
Solidary distribution between funds*	**1**	1/7							
Clear responsibility and accountability	**5**		1/7	3/6					1/6
Named coordinator	**7**		1/7	4/6		1/7	1/7		
Continuity of care	**9**		2/7	2/6	3/6	1/7		1/6	
Collaboration across sectors*	**5**			4/6					1/6
Coordination between primary care and hospitals*	**1**							1/6	
Prevent and avoid going to ER*	**2**							2/6	
Administrative barriers*	**1**		1/7						
Transferring of responsibility*	**1**		1/7						
Transfer-care*	**2**			1/6		1/7			
After-care*	**2**			1/6		1/7			
Systematic operating procedures in care*	**3**				3/6				
Utilisation of EMRs*	**7**			3/6	2/6	2/7			
Careful use of patient files / privacy	**2**			2/6					
Confidence in knowledge and skills in providers	**10**	1/7	2/7	3/6	2/6	1/7		1/6	
Education of health professionals*	**3**		3/7						
Importance and function of centres of excellence*	**1**		1/7						
Broader specialisations*	**1**						1/7		

- **Box S.3** continued

	# / 58	**Austria**	**Croatia**	**Germany[1]**	**Hungary**	**Netherlands**	**Norway**	**Spain**	**UK**
Access & availability									
Physical access	**5**		1/7	1/6		2/7		1/6	
Timely access* (i.e., short waiting times)	**9**		2/7	1/6	2/6	1/7			4/6
Enough time* (provider for patient)	**8**	4/7			1/6	1/7		2/6	
Being seen by specialist when needed*	**2**							2/6	
Geographical access	**3**		1/7					1/6	1/6
Availability of treatment*	**3**		3/7						
Home care (available)	**1**			1/6					
Transport*	**1**			1/6					
*Avoid unnecessary treatment**	**2**	2/7							
Pro-active, prevention-oriented, care	**13**		1/7	1/6	4/6	4/7			3/6
Patient-education and support (groups)									
Integration of self-help groups*	**3**	3/7							
Importance of associations of patients*	**1**		1/7						
Good trainings offered*	**1**	1/7							
Medication help*	**5**			1/6		4/7			
Self-management support*	**1**					1/7			
Good info for patients on health	**1**						1/7		
Family support*	**3**		3/7						
*Bureaucratic hurdles**	**5**			4/6		1/7			
Physical surroundings									
Material conditions in institutions*	**1**		1/7						
Macro-level									

- **Box S.3** continued

	# / 58	**Austria**	**Croatia**	**Germany**[1]	**Hungary**	**Netherlands**	**Norway**	**Spain**	**UK**
Non-cooperation with health policy makers*	**2**		2/7						
Importance of media pressure	**2**		2/7						
Learning from different countries' experiences*	**1**	1/7							
Overall satisfaction with care process	**8**	1/7		4/6	2/6			1/6	
Costs									
Loss of income									
Loss of income informal caregiver	**3**		1/7					2/6	
Loss of income patient	**7**	1/7	3/7		1/6			1/6	1/6
Overall costs in relation to income*	**1**					1/7			
Service and support coverage	**6**	2/7	1/7	1/6	1/6		1/7		
*Need for cost-benefit analysis**	**2**		2/7						
*Money follows the patient** (bundled type payment that crosses all care level-borders)	**1**		1/7						
Travel and parking costs	**2**				1/6				1/6

*Note: 1 = Group split into 2, so each time # out of 6 persons, each group discussed either health/well-being and costs or care and costs. Btw = between, Profs = professional care providers, EMRs = Electronic Medical Records, ER = Emergency Room. * Concepts/phrases that were novel and written on cards based on part 1 of the focus groups, and not on the basis of the a priori literature search.*

● **Box S.4** Criteria used to come to a core set of outcomes to evaluate integrated care programmes

- **Cover the Triple Aim**: 1) improving population health/well-being and 2) patient experience, and 3) reducing cost (growth);
- **Measurable at an individual level** using PROMS and PREMS (this only applies to the first and second of the three aims);
- **Relevant for multi-morbidity** in different contexts and population groups;
- **Relevant for the 5P stakeholders: p**atients (persons with multi-morbidity), **p**artners (informal caregivers), **p**rofessionals, **p**ayers, **p**olicy makers.
- **Relevant across the** 17 selected **promising integrated care programmes**;
- **Timing of intervention effects**: the intervention effect (i.e., the impact of a programme on the outcome) in newly enrolled patients is likely to start occurring within the SELFIE evaluation period (6-18 months);
- The **outcomes are not redundant**, meaning that they are all necessary and not superfluous (e.g., one outcome does not overlap much with another outcome);
- **Preference independence**, i.e., the weight of one outcome can be elicited independently from the performance of another outcome;
- **Feasible to measure** performance.

Chapter

3

Heterogeneity In Preferences For Outcomes Of Integrated Care For Persons With Multiple Chronic Diseases: A Latent Class Analysis Of A Discrete Choice Experiment

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ABSTRACT

Purpose: For an integrated care programme to be successful, preferences of the stakeholders involved should be aligned. The aim of this study is to investigate to which extent outcomes beyond health are valued and to study the heterogeneity of preferences of those involved in integrated care.

Methods: A discrete choice experiment (DCE) was conducted to elicit preferences for eight Triple Aim outcomes, i.e., physical functioning, psychological well-being, social relationships & participation, enjoyment of life, resilience, person-centeredness, continuity of care and total health and social care costs. Stakeholders were recruited among Dutch persons with multi-morbidity, informal caregivers, professionals, payers, and policymakers. A Bayesian mixed-logit model was used to analyse the data. Subsequently, a latent class analysis was performed to identify stakeholders with similar preferences.

Results: 739 stakeholders completed the DCE. Enjoyment of life was perceived as the most important outcome (relative importance: 0.221) across stakeholders, while total health and social care costs were perceived as least important (0.063). The latent class analysis identified four classes. The first class (19.9%) put most weight on experience with care outcomes. The second class (39%) favoured enjoyment of life. The third class (18%), focused relatively more on physical health. The fourth class (24%) had the least consistent preferences.

Conclusion: This study has highlighted the heterogeneity in views of stakeholders in integrated care on what is important in health(care) for persons with multi-morbidity. To accurately value integrated care a variety of outcomes beyond health – e.g., enjoyment of life and experience with care – should be taken into account.

INTRODUCTION

Integrated care programmes that focus on multi-morbidity often include a package of complex and multifaceted interventions that have multiple aims [1][2]. Such aims include improved population health, better patient experience, cost reduction (known as the Triple Aim [3]), and better experience of providing care (known as the Quadruple Aim [4]). Therefore, evaluations of such models of care require the measurement of a broad spectrum of outcomes that go beyond traditional health outcomes like health-related quality of life and longevity [5].

However, not all aims are expected to have the same importance to the different stakeholders involved in the design, provision, financing, and receipt of integrated care for persons with multi-morbidity. For example, patients may assign higher importance to experience with care than clinicians with overburdened workloads, while payers may be more sensitive to costs than other stakeholders. Discordance in preferences complicates the decision-making process [6]. International experience and scientific evidence show that the success of integrated care models is highly dependent on the alignment of stakeholder preferences for the model's aims and achievements [7-9]. Hence, it is important to elicit their preferences and take them into account when designing and assessing integrated care for multi-morbidity [9].

A common technique to measure preferences in healthcare delivery is a discrete choice experiment (DCE), in which respondents are asked to make a number of choices between two hypothetical options characterised by attributes with differing levels [10]. A DCE forces respondents to make trade-offs between multiple elements or aims of a health care intervention [11]. Especially in integrated care this is important as interventions focus on improving outcomes beyond the quality-adjusted life year (QALY). An advantage of a DCE is that, by making patient-preferences so explicit, it makes it possible to incorporate them in decision-making [12]. To study heterogeneity in choices, one can use a latent class analysis to identify underlying subgroups of respondents with similar preferences and characterised by background characteristics. This information can be used to better understand differences in preferences between stakeholders and further align them.

The aim of this study was (1) to investigate to which extent outcomes beyond health are valued and (2) to study the heterogeneity of preferences for outcome measures of integrated care among stakeholders involved in integrated care. The outcome measures included in the preference study were physical functioning, psychological well-being, social relationships & participation, enjoyment of life, resilience, person-centeredness, continuity of care, and total health and social care costs. Respondents were recruited among persons with multi-morbidity, partners & other informal caregivers, professionals, payers, and policymakers. This is the first DCE study including such a wide variety of outcomes measures relevant to integrated care and such a diversity of stakeholders involved in integrated care.

METHODS

Context of the DCE

This study took place in the context of the EU-funded SELFIE2020 project, in which we aimed to elicit preferences for outcome measures of integrated care that could be used in Multi-Criteria Decision Analysis [13], see **box 1**. In the current study, preference data from Dutch stakeholders involved in integrated care were used.

Box 1

SELFIE was a Horizon2020 funded EU project that aimed to contribute to the improvement of person-centred care for persons with multi-morbidity by proposing evidence-based, economically sustainable, integrated care programmes that stimulate cooperation across health and social care and are supported by appropriate financing and payment schemes. More specifically, SELFIE aimed to:

- Develop a taxonomy of promising integrated care programmes for persons with multi-morbidity;
- Provide evidence-based advice on matching financing/payment schemes with adequate incentives to implement integrated care;
- Provide empirical evidence of the impact of promising integrated care on a wide range of outcomes using Multi-Criteria Decision Analysis;
- Develop implementation and change strategies tailored to different care settings and contexts in Europe, especially Central and Eastern Europe.

Seventeen promising integrated care programmes for persons with multi-morbidity are being evaluated in SELFIE using MCDA and a common set of core outcomes as well as programme-type specific outcomes. The latter depend on whether a programme is i) a population health management programme, ii) a programme targeting frail elderly, iii) a programme targeting persons with problems in multiple life domains, or iv) an oncology or palliative care programme.

The SELFIE consortium included eight organisations in the following countries: the Netherlands (coordinator) (NL), Austria (AT), Croatia (HR), Germany (DE), Hungary (HU), Norway (NO), Spain (ES), and the United Kingdom (UK). (www.SELFIE2020.eu) [Grant Agreement No 634288]

Attributes and levels

The development of attributes and attribute levels (see **Table 1**) consisted of two steps. First, a longlist with potentially relevant attributes was composed using four methods: 1) a literature review of outcome measures used in (integrated) care, 2) national workshops with patients, informal caregivers, professionals, payers and policymakers in the eight countries in the SELFIE project to discuss outcomes of integrated care, 3) eight focus groups with individuals with multi-morbidity to discuss what outcomes of integrated care matter to them [14], and 4) a review of outcomes being used in the 17 integrated care programmes in Europe that were evaluated in the SELFIE project. The second step was to shorten the list, a process that was guided by multiple criteria, including relevance to multi-morbidity in different contexts and population groups, non-redundancy, operationality, and preference independence [13]. The levels to describe the attributes were defined such that they represent the full range of the scale from worst to best, with an intermediate level in between. The wording of the levels was based on validated questionnaires that are used to measure these outcomes in empirical research [13]. For costs we used estimates of the mean total health and social care costs for people with multimorbidity in the Netherlands (middle level), which we increased and decreased by 20%.

Design

Given the large number of attributes, in combination with the three possible levels for each, a full factorial design that includes all 6561 possible alternatives (i.e., 3^8: 8 attributes with 3 attribute levels), would not be feasible. To reduce this set of combinations to a manageable number, we used specialised software to select the most informative combinations of attribute levels per choice question, using Bayesian design algorithms that maximise the D-efficiency for a pre-specified conditional logit main-effects model [15-17]. Maximising the D-efficiency involves minimising the confidence sphere around the complete set of model parameters in this logit model. Priors for the weights of the attribute-levels, as required for an efficient optimisation approach, were obtained from literature [18]. To further improve the efficiency of the parameter estimates obtained from the DCE, the overall DCE design comprised ten different sub-designs. This means that instead of using 1 design for all respondents, we constructed 10 different sub-designs, and each respondent is only asked to complete one, randomly chosen, sub-design that consists of a pre-specified number of 18 choice tasks [15]. The informative priors from the literature were updated using the answers of the first 50 respondents from each stakeholder group, to create a more efficient DCE design for the remaining respondents in the stakeholder group.

- **Table 1.** Attributes and levels

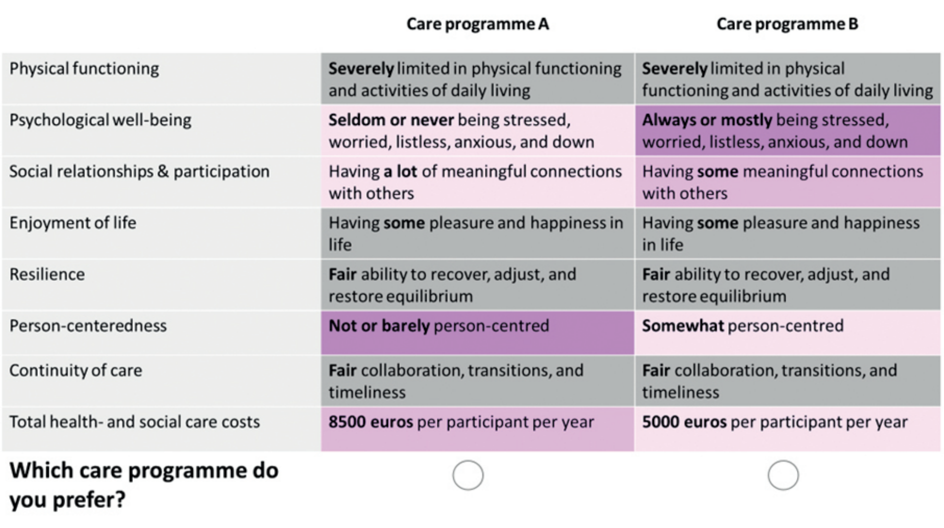
Attributes (outcome measures)	Levels
Physical functioning	**1. Severely** limited in physical functioning and activities of daily living **2. Moderately** limited in physical functioning and activities of daily living **3. Hardly or not at all** limited in physical functioning and activities of daily living
Psychological well-being	**1. Always or mostly** stressed, worried, listless, anxious, and down **2. Regularly** stressed, worried, listless, anxious, and down **3. Seldom or never** stressed, worried, listless, anxious, and down
Social relationships and participation	**1. No or barely any** meaningful connections with others **2. Some** meaningful connections with others **3. A lot** of meaningful connections with others
Enjoyment of life	**1. No or barely any** pleasure and happiness in life **2. Some** pleasure and happiness in life **3. A lot** of pleasure and happiness in life
Resilience	**1. Poor** ability to recover, adjust, and restore balance **2. Fair** ability to recover, adjust, and restore balance **3. Good** ability to recover, adjust, and restore balance
Person-centeredness	**1. Not or barely** person-centred **2. Somewhat** person-centred **3. Highly** person-centred
Continuity of care	**1. Poor** collaboration, transitions, and timeliness **2. Fair** collaboration, transitions, and timeliness **3. Good** collaboration, transitions, and timeliness
Total health- and social care costs	**1. €8500** per participant per year **2. €7000** per participant per year **3. €5500** per participant per year

When scanning the subsets of the full-choice design to find a D-optimal design we imposed two design constraints to reduce the complexity and to avoid unrealistic choice tasks. First, the highest level of enjoyment of life and lowest level of psychological well-being, and vice versa, were never combined within a single choice option, i.e., ‘Seldom or never stressed, worried, listless, anxious, and down’ (highest level of psychological well-being), and ‘No or barely any pleasure and happiness in life’ (lowest level of enjoyment of life) could never be part of the same programme description. Second, in each choice task either three or four attributes needed to have the same level for each alternative to reduce overall complexity and improve response efficiency [19].

Questionnaire

The DCE questionnaire was self-administered and web-based. After informed consent, the meaning of the attributes and levels was explained. Each choice task described two alternatives with eight attributes of varying levels. These two alternatives represented two integrated care programmes with different outcome-profiles. These were labelled ‘Care programme A’ and ‘Care programme B’ (See **Figure 1** for an example choice task). Respondents were asked to complete two ‘warm-up’ DCE choice-tasks before the main choice tasks. This familiarised respondents with the attributes and levels and prepared them for the full set of 18 choice tasks. The choice tasks were presented in three groups of six choice tasks each, with a few general demographic or health-related questions in between to reduce the repetitive nature of the choice tasks. The questionnaire concluded with debriefing questions related to the stakeholder perspectives the respondents identify themselves with and the ease of understanding and completion of the choice tasks.

- **Figure 1.** Example choice task DCE



	Care programme A	**Care programme B**
Physical functioning	**Severely** limited in physical functioning and activities of daily living	**Severely** limited in physical functioning and activities of daily living
Psychological well-being	**Seldom or never** being stressed, worried, listless, anxious, and down	**Always or mostly** being stressed, worried, listless, anxious, and down
Social relationships & participation	Having **a lot** of meaningful connections with others	Having **some** meaningful connections with others
Enjoyment of life	Having **some** pleasure and happiness in life	Having **some** pleasure and happiness in life
Resilience	**Fair** ability to recover, adjust, and restore equilibrium	**Fair** ability to recover, adjust, and restore equilibrium
Person-centeredness	**Not or barely** person-centred	**Somewhat** person-centred
Continuity of care	**Fair** collaboration, transitions, and timeliness	**Fair** collaboration, transitions, and timeliness
Total health- and social care costs	**8500 euros** per participant per year	**5000 euros** per participant per year
Which care programme do you prefer?	○	○

The DCE questionnaire was pilot tested with six patients, including think aloud sessions to see if there were elements that needed clarification. After the pilot study small changes were made in the visual design of the study to enhance the clarity of the questionnaire.

Subject recruitment and data collection

To recruit a representative group of different stakeholders involved in integrated care, we aimed to recruit 750 respondents among persons with multi-morbidity (n=150), informal caregivers of persons with multi-morbidity (n=150), professionals (n=150), payers (n=150) and policymakers (n=150). In July 2017, members of an online marketing research panel who were persons with multi-morbidity, informal caregivers, or professional care providers were invited to complete the questionnaire. Payers and policymakers were invited via the same panel organisation, but since it was difficult to reach 150 respondents in these groups, recruitment was supplemented by personal invitations of payers and policymakers in the network of the researchers of the SELFIE project, followed by snowballing. Between July 2017 and July 2018, we approached healthcare payers such as health insurance companies and departments of municipalities responsible for paying social care. To include policymakers, we invited public servants working at the ministry of health, provincial or local governments, official governmental advisory bodies, mayors, aldermen, and city councillors with health and/or social care in their portfolio. Each participant was asked to confirm that they belonged to their assigned stakeholder group and to respond from that perspective. They were also invited to indicate one or more other stakeholder group(s) that they felt they belonged to as well. It was made impossible to fill in the questionnaire using a mobile phone or tablet as the choice task would not be fully visible.

Statistical analysis

First, a Bayesian mixed logit model (MIXL), using diffuse priors for the mean values of the random coefficients, was used to analyse the data of all respondents simultaneously. This model allowed all utility coefficients to be randomly distributed and estimated a full covariance matrix among them. A burn-in phase of 10,000 Markov Chain Monte Carlo (MCMC) draws was used, followed by 30,000 draws to reliably approximate the posterior. The relative importance of each attribute (i.e., outcome measure of integrated care) was based on the coefficient of its best level (level 3) divided by the sum of all best attribute levels.

Second, a latent class model was used to model heterogeneity across individuals with a discrete distribution over a set of classes, and individuals were sorted into a set of classes based on their observed choice data [20]. Latent class analysis is an extension of the standard logit model and is used to identify unobserved groups of similar individuals (latent classes) with homogeneous preferences based on observed variables. These distinct groups can possess (widely) different preferences regarding integrated care. Furthermore, latent class modelling is probabilistic, which means that respondents are allocated to the group they are most likely to be a member of. Subsequently, posterior analysis can be used to describe differences in characteristics across groups. Initially, we compared 2 to 9 class solutions for the best statistical fit based on the Bayesian Information Criterion (BIC) and Consistent Aikake Information Criterion (CAIC), with a lower value implying a better fit [21]. We also considered theoretical interpretability and the size of the classes to see if another number of classes would be more logical based on the observed variables. For each respondent, the posterior probability that (s)he belongs to each latent class was calculated and each respondent was assigned to the class with the highest probability. Class membership was based solely on estimated preferences from the DCE. To assess whether differences between classes were significant, we conducted chi-square tests (categorical variables), one-way ANOVA tests (continuous variables) and Kruskal Wallis tests (non-parametric test for continuous variables).

Third, as the observed preference heterogeneity could be related to stakeholders having multiple roles, we investigated which other perspectives the stakeholders identified themselves with, in a descriptive analysis.

Analyses were performed in Stata 16.0, using the bayesmixedlogit module specified with Metropolis-within-Gibbs sampling and default (uninformative) priors for the MIXL model and the lclogit procedure for the latent class models.

RESULTS

Study population

There were 935 persons that started the questionnaire and gave informed consent, of which 739 (79%) finished all DCE scenarios. The mean time to complete the questionnaire was approximately 20 minutes. 705 respondents spent at least 5 minutes filling in the questionnaire and all further analyses are performed on this group.

Table 2 summarises the characteristics of the respondents. Their mean age was 49.6 years, 54.2% was female, and the majority (73.1%) of respondents was employed. The respondents' highest attained educational level was relatively high. 31% of the respondents reported no health problems.

- **Table 2.** Respondent characteristics

Participation	
Started questionnaire and gave informed consent **n**	935
Finished all DCE scenario's **n**	739
Mean time to completion **(SD)**	19.90 (12.53)
5-90 minutes **n**	705
Demographics (N=705)	
Mean age **(SD)**	49.59 (14.05)
Median age **(min-max)**	51 (21-88)
Gender – female **n (%)**	382 (54)
Educational level **n (%)**	
1. Low	25 (4)
2. Medium	188 (27)
3. High	492 (70)
Work status **n (%)**	
1. Paid job	515 (73)
2. Volunteer work	136 (19)
3. Retired / pre-pension	103 (15)
4. [Partially] Work disabled	44 (6)
5. Looking for a job	19 (3)
6. Do not have paid job	12 (2)
7. Housewife/househusband	63 (9)
8. Student	36 (5)
Health characteristics	
General health **n (%)**	
1. Excellent	99 (14)
2. Very good	166 (24)
3. Good	282 (40)
4. Fair	133 (19)
5. Poor	25 (4)
Mean general health **(SD)**	2.74 (1.03)

- **Table 2.** Continued.

Health conditions (top 10 most frequent) n (%)	
1. Depression, anxiety or emotional difficulties	86 (12)
2. Colon problem, irritable bowel or colitis	82 (12)
3. Chronic back pain or sciatica	67 (10)
4. Diabetes	66 (9)
5. Osteoarthritis (not rheumatoid arthritis)	60 (9)
6. Asthma	42 (6)
7. Rheumatoid arthritis	42 (6)
8. Chronic bronchitis, COPD or emphysema	41 (6)
9. Heart disease, angina, heart attack, bypass surgery or angioplasty	40 (6)
10. Stomach problem, ulcer, gastritis or reflux	38 (5)
No health problems	217 (31)
Other health problems	102 (14)
I prefer not to answer	27 (4)
Mean number of health problems **(SD)**	1.79 (2.08)
Stakeholder group n (%)	
1. Person with multi-morbidity	158 (22)
2. Informal caregiver	152 (22)
3. Professional	148 (21)
4. Payer	102 (14)
5. Policymaker	145 (21)
Difficulty DCE choice tasks n (%)	
1. Very easy	25 (4)
2. Easy	181 (26)
3. Not too easy, not too difficult	291 (42)
4. Difficult	175 (25)
5. Very difficult	18 (3)
Mean difficulty **(SD)**	2.97 (0.87)

DCE preferences across all stakeholders

The results of the Bayesian MIXL (**Table 3**) showed that all attribute-levels differed from level 1. The attribute levels had the expected positive sign, and the coefficients of level 3 were always larger than those of level 2. This means that level 2 and level 3 were valued higher than level 1 and the level 3 was valued higher than level 2. On average, the respondents assigned the highest relative importance to enjoyment of life, followed by psychological well-being, and resilience (**Figure 2**). The least important outcome was total health and social care costs. However, the standard deviations of all attribute (levels) indicated a wide variation in preferences among respondents (**Table 3**).

- **Table 3.** Attribute-level coefficients of the Bayesian MIXL model

Attribute (i.e., outcome measure)	Level	Mean	95% Credible interval	Standard deviation	95% Credible interval
Physical functioning	2	2.29	2.03 - 2.55	2.13	1.86 – 2.41
	3	3.32	2.97 - 3.67	3.13	2.78 – 3.51
Psychological well-being	2	2.02	1.79 - 2.25	1.79	1.55 – 2.04
	3	4.04	3.65 - 4.44	3.54	3.16 – 3.95
Social relationships & participation	2	1.74	1.53 - 1.95	1.54	1.33 – 1.77
	3	2.43	2.17 - 2.69	2.19	1.92 – 2.47
Enjoyment of life	2	3.61	3.30 - 3.92	2.30	2.03 – 2.60
	3	5.57	5.11 - 6.04	3.75	3.35 – 4.16
Resilience	2	2.54	2.31 - 2.77	1.61	1.39 – 1.86
	3	3.44	3.14 - 3.74	2.19	1.92 – 2.50
Person-centeredness	2	1.26	1.08 - 1.45	1.27	1.06 – 1.47
	3	2.09	1.85 - 2.32	1.91	1.67 – 2.17
Continuity of care	2	2.05	1.83 - 2.27	1.67	1.43 – 1.92
	3	2.69	2.43 - 2.95	2.12	1.83 – 2.43
Total health- and social care costs	2	0.74	0.59 - 0.90	1.13	0.96 – 1.33
	3	1.58	1.37 - 1.80	1.87	1.63 – 2.13

Note: The coefficients represent the respondent's preferences for the various attributes and their levels. Each attribute consisted of 3 levels, with level 3 as best performing level. A higher coefficient reflects a higher preference.

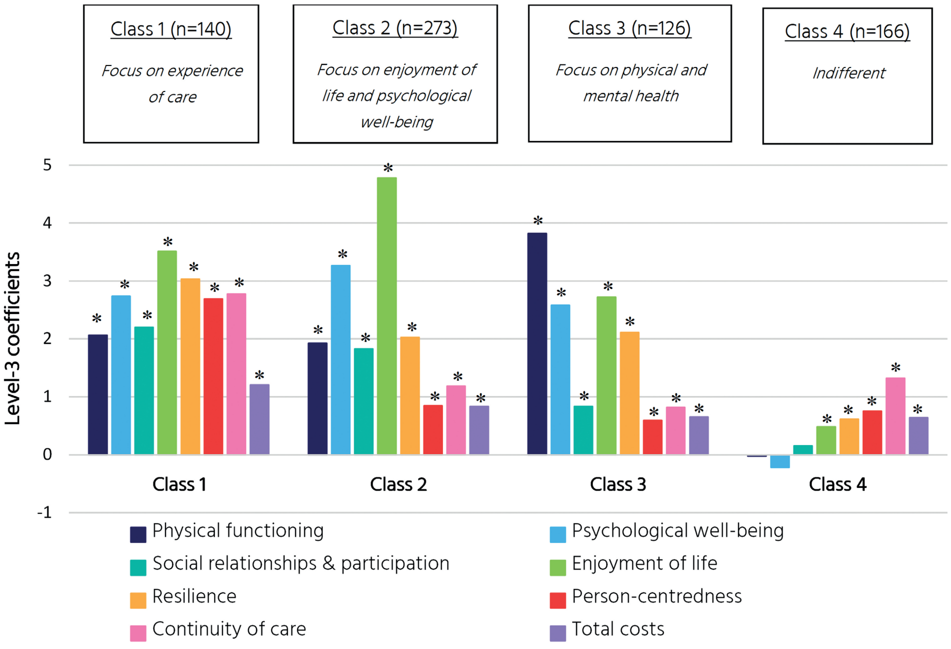
Figure 2. Relative importance of the outcome measures

** All relative importance weights sum up to 1. The relative importance of each outcome measure was based on the coefficient of its attribute level-3 divided by the sum of all level-3 coefficients.*

Grouping stakeholders with similar preferences

Based on the information criteria (BIC and CAIC), the latent class model with four classes provided the best model fit. Based on the class probabilities 20% (n=140) of all respondents was assigned to class 1, 39% (n=273) to class 2, 18% (n=126) to class 3, and 24% (n=166) to class 4. The average of the respondents' maximum posterior class membership probabilities was 0.82 (SD=0.17, median=0.87), varying from 0.74 for class 1 and 0.89 for class 4. **Figure 3** presents the class-specific preference coefficients. The estimates in class 1, 2 and 3 had the expected direction, i.e., respondents preferred a higher level of each outcome. In class 4 the preferences for physical functioning, psychological well-being and social relationships & participation were not statistically significant. A table with all coefficients, including standard errors and p-values, is presented in **Appendix 1**.

Figure 3. Results latent class analysis: coefficients of attribute-level 3



** Significant preference within the class (P<0.05)*

Compared to the other classes, class 1 respondents had the highest estimates for the experience with care outcomes (continuity of care and person-centeredness). In class 2, respondents assigned a relatively higher weight to enjoyment of life, followed by psychological well-being, than the other classes. Respondents in class 3 had a stronger preference for physical health than the other classes, followed by three outcomes related to mental health i.e., psychological well-being, enjoyment of life and resilience. The coefficients in class 4 were overall quite small, which indicates that the preferences were less consistent than in the other classes.

Characteristics of stakeholders with similar preferences

Table 4 presents the background characteristics of stakeholders in the four latent classes. All classes included representatives from all primary stakeholder groups, although professionals were overrepresented in class 1 (26%), followed by policymakers (25%). Class 1 had the highest share of females (62%). Also, health was best in this class compared to the other classes based on both self-perceived general health (mean 2.54 and 44% of respondents who answered “excellent” or “very good”) and the mean number of health conditions (1.55).

- **Table 4.** Class-specific respondent characteristics

	Class 1 n=140	Class 2 n=273	Class 3 n=126	Class 4 n=166	
Participation					**P-value**
Time to completion - mean (SD)	20.36 (13.61)	20.77 (13.02)	19.84 (11.80)	18.16 (11.17)	0.000
Time to completion - median (min-max)	17.31 (6.97-85.07)	17.35 (5.13-85.07)	16.58 (5.75-77.95)	15.74 (5.13-83.38)	
Demographics – n (%)					**P-value**
Age - mean (SD)	48.59 (14)	47.63 (14)	49.56 (14)	53.67 (14)	0.000
Gender (female)	87 (62)	156 (57)	52 (41)	87 (52)	0.000
Educational level					0.000
1. Low	5 (4)	9 (3)	2 (2)	9 (5)	
2. Medium	32 (23)	59 (22)	33 (26)	64 (39)	
3. High	103 (74)	205 (75)	91 (72)	93 (56)	
Work status					0.000
1. Paid job	102 (73)	216 (79)	99 (79)	98 (59)	
2. Volunteer work	30 (21)	53 (19)	16 (13)	37 (22)	
3. Retired / pre-pension	23 (16)	30 (11)	15 (12)	35 (21)	
4. [Partially] Work disabled	8 (6)	12 (4)	10 (8)	14 (8)	
5. Looking for a job	3 (2)	7 (3)	3 (2)	6 (4)	
6. Do not have paid job	5 (4)	4 (1)	2 (2)	1 (1)	
7. Housewife/ househusband	11 (8)	25 (9)	6 (5)	21 (13)	
8. Student	7 (5)	17 (6)	5 (4)	7 (4)	
Health characteristics					**P-value**
General health **n (%)**					0.000
1. Excellent	24 (17)	39 (14)	20 (16)	16 (10)	
2. Very good	38 (27)	68 (25)	30 (24)	30 (18)	
3. Good	58 (41)	113 (41)	43 (34)	68 (41)	
4. Fair	18 (13)	46 (17)	25 (20)	44 (27)	
5. Poor	2 (1)	7 (3)	8 (6)	8 (5)	
Mean general health **(SD)**	2.54 (1)	2.69 (1)	2.77 (1)	2.99 (1)	0.000

Table 4. Continued.

	Class 1 n=140	Class 2 n=273	Class 3 n=126	Class 4 n=166
Health conditions n (%)				
1. Colon problem, irritable bowel or colitis	15 (11)	36 (13)	15 (12)	16 (10)
2. Depression, anxiety or emotional difficulties	10 (7)	32 (12)	16 (13)	28 (17)
3. Chronic back pain or sciatica	14 (10)	20 (7)	14 (11)	19 (11)
4. Osteoarthritis (not rheumatoid arthritis)	14 (10)	20 (7)	7 (6)	19 (11)
5. Chronic bronchitis, COPD or emphysema	7 (5)	17 (6)	7 (6)	10 (6)
6. Diabetes	5 (4)	16 (6)	15 (12)	30 (18)
7. Heart disease, angina (chest pain from heart problem), heart attack, bypass surgery or angioplasty	8 (6)	16 (6)	9 (7)	7 (4)
8. Rheumatoid arthritis	8 (6)	15 (5)	6 (5)	13 (8)
9. Stomach problem, ulcer, gastritis or reflux	9 (6)	15 (5)	2 (2)	12 (7)
10. Asthma	6 (4)	14 (5)	11 (9)	11 (7)
11. Poor circulation in your legs	8 (6)	14 (5)	8 (6)	9 (5)
12. Thyroid disorder	6 (4)	14 (5)	6 (5)	7 (4)
13. Cancer during the past five years	3 (2)	5 (2)	5 (4)	11 (7)
14. Congestive heart failure	3 (2)	2 (1)	4 (3)	9 (5)
No health problems	49 (35)	93 (34)	38 (30)	37 (22)
Other health problems	20 (14)	41 (15)	12 (10)	29 (17)
I prefer not to answer	4 (3)	10 (4)	3 (2)	10 (6)

- **Table 4.** Continued.

	Class 1 n=140	Class 2 n=273	Class 3 n=126	Class 4 n=166	
Multi-morbidity n (%)					**P-value**
1 health problem	29 (21)	53 (20)	23 (19)	35 (22)	
2 health problems	16 (12)	38 (14)	17 (14)	17 (11)	
3 or more health problems	32 (24)	67 (25)	43 (35)	57 (37)	
Mean number of health problems **(SD)**	1.55 (2.21)	1.62 (1.88)	1.85 (1.84)	2.23 (2.41)	0.000
Median number of health problems **(min-max)**	1 (0-15)	1 (0-7)	1 (0-9)	1 (0-11)	
Stakeholder group n (%)					**P-value**
Stakeholder group					0.000
1. Person with multi-morbidity	27 (19)	57 (21)	36 (29)	38 (23)	
2. Informal caregiver	24 (17)	51 (19)	21 (17)	56 (34)	
3. Professional	37 (26)	62 (23)	22 (17)	27 (16)	
4. Payer	17 (12)	50 (18)	18 (14)	17 (10)	
5. Policymaker	35 (25)	53 (19)	29 (23)	28 (17)	
Number of additional perspectives chosen by respondent n (%)					**P-value**
0 additional perspectives	64 (47)	139 (51)	63 (50)	68 (43)	
1 additional perspective	51 (37)	101 (37)	46 (37)	68 (43)	
2 or more additional perspectives	22 (16)	30 (11)	16 (13)	22 (14)	
Mean number of additional perspectives chosen (**SD**)	0.74 (0.0.84)	0.63 (0.76)	0.65 (0.77)	0.73 (0.77)	0.000
Additional perspective n (%)*					**P-value**
Additional perspective					0.000
1. Person with multi-morbidity	21 (15)	37 (14)	17 (13)	28 (17)	

Table 4. Continued.

	Class 1 n=140	Class 2 n=273	Class 3 n=126	Class 4 n=166	
2. Informal caregiver	30 (22)	45 (17)	20 (16)	33 (21)	
3. Professional	16 (12)	26 (10)	14 (11)	16 (10)	
4. Payer	14 (10)	20 (7)	15 (12)	21 (13)	
5. Policymaker	20 (15)	41 (15)	15 (12)	18 (11)	
Difficulty n (%)					**P-value**
Difficulty					0.000
1. Very easy	1 (1)	10 (4)	4 (3)	10 (6)	
2. Easy	35 (26)	71 (26)	34 (27)	41 (26)	
3. Not too easy, not too difficult	62 (45)	113 (42)	57 (46)	59 (37)	
4. Difficult	37 (27)	71 (26)	25 (20)	42 (27)	
5. Very difficult	2 (1)	5 (2)	5 (4)	6 (4)	

** This percentage is based on the number of respondents that chose a certain perspective divided by the total number of respondents in the respective class, i.e., the numbers do not vertically add up to the n of the class, as some respondents chose no additional perspective.*

In class 2 the stakeholder groups were quite evenly distributed, with somewhat more professionals (23%). This class consisted of respondents with the lowest mean age (47.6 years). Furthermore, the educational level and employment rate were the highest in this class. 75% of the respondents had a high educational level and 79% currently had a paid job.

Persons with multi-morbidity were more frequently a member of class 3 (29%). This class, which predominantly consisted of males (59%), also had a worse health status than respondents in class 1 and 2.

Class 4 had the highest share of informal caregivers (34%). The respondents in this class were on average older (53.7 years), lower educated, in worse general health (31% “fair” or “poor” health) and had more health problems (mean 2.23 health problems) than respondents in the other classes. Of the respondents in this class, 30% found the questionnaire difficult or very difficult, although the time to completion was the fastest of all classes (18.2 minutes).

Multiple perspectives per stakeholder

Of the respondents, 48% chose no additional stakeholder perspective they identify themselves with. 39% of the respondents only selected one additional perspective, 13% identified with two or more additional perspectives. In the entire sample, 18% of the persons with multi-morbidity identified themselves as informal caregiver (**Table 5**). Of the informal caregivers, 29% identified themselves as person with multi-morbidity. 30% of the professionals identified themselves with the perspective of informal caregiver. 59% of the payers viewed themselves as policymaker. Of the policymakers, 24% selected informal caregiver as additional perspective.

When comparing the four classes (Table 4), class 4 had the lowest percentage of respondents that chose no additional perspective (43%). In all four classes 'Informal caregiver' was the most frequent additional perspective and in classes 1, 3 and 4 this was followed by person with multi-morbidity. There were no marked differences between the classes in the additional perspectives that were chosen.

- **Table 5.** Overlap in perspectives of respondents

		Stakeholders identifying as belonging to multiple groups					
Assigned stakeholder group	**n**	1. Person with multi-morbidity	2. Informal caregiver	3. Professional	4. Payer	5. Policy-maker	n (%)* that chose at least 1 additional perspective
1. Person with multi-morbidity	158 (22%)		**29 (18%)**	**10 (6%)**	**14 (9%)**	**13 (8%)**	52 (33%)
2. Informal caregiver	152 (22%)	**44 (29%)**		**17 (11%)**	**15 (10%)**	**8 (5%)**	70 (46%)
3. Professional	148 (21%)	**22 (15%)**	**45 (30%)**		**13 (9%)**	**13 (9%)**	73 (49%)
4. Payer	102 (15%)	**14 (14%)**	**20 (20%)**	**16 (16%)**		**60 (59%)**	79 (77%)
5. Policymaker	145 (21%)	**23 (16%)**	**34 (24%)**	**29 (20%)**	**28 (19%)**		82 (57%)
Total	705	103 (15%)	128 (18%)	72 (10%)	70 (10%)	94 (14%)	

** Note: The numbers do not add up to this total because respondents were allowed to select more than one additional stakeholder perspective*

DISCUSSION

Interpretation of the main findings

This study investigated stakeholders' preferences for outcomes of integrated care for persons with multi-morbidity using a DCE. Based on the mixed logit model results of the pooled data, which showed that all attribute levels were statistically different from 1, it was established that all outcome measures and all levels of the outcomes measures influenced stakeholders' choices. This means that stakeholders took all outcome measures into account when deciding upon which care programme was preferred. Enjoyment of life, psychological well-being and resilience were deemed most important and total health and social care costs least important, but there was a lot of variation in preferences.

When divided into four classes using latent class analysis, we could identify a class that assigned a relatively higher weight to the two experience with care outcomes, i.e., continuity of care and person-centeredness, a class that emphasised the importance of enjoyment of life and psychological well-being, a class that was more focused on physical functioning and a class with inconsistent preferences. Each of the classes included persons with multi-morbidity as well as informal caregivers, professionals, payers and policy makers, suggesting that differences between the classes were not particularly driven by stakeholder group. One of the reasons that differences in preferences cannot be directly related to stakeholder perspective might be that respondents obviously have multiple roles, as was clearly shown by their self-reported additional stakeholder perspectives. Many informal caregivers, payers and policy makers were patients themselves.

Nevertheless, some stakeholders were overrepresented in some classes. The class that focused more on experience with care (class 1) included relatively more professionals and policy makers, i.e., stakeholders that are used to monitoring process outcomes as indicators of the quality of care. In the Dutch context, where patients are free to choose between care providers, professionals are incentivised to improve patient's experience and satisfaction with their services to increase their market share. This is emphasised by payers who consider quality indicators when contracting providers. The class that focused on physical and mental health (class 3) had the highest share of persons with multi-morbidity, resulting in the highest share of persons reported having more than one health problem. A likely explanation is that people with immediate concerns about their health prefer outcomes related to these domains in contrast to experience with care or cost outcomes. In the class with less consistent preferences (class 4), persons with multi-morbidity and informal caregivers were overrepresented. Respondents in this class found the questionnaire difficult, which has likely contributed to the inconsistency. The fact that they had the shortest completion time might illustrate this difficulty.

The largest class (class 2) with approximately 39% of the sample, consisted of respondents that put much weight on enjoyment of life. The respondents were quite evenly distributed over the stakeholders. The respondents in this class were relatively younger, higher educated, healthier or more likely to have a paid job than respondents in class 3 and 4. Their lesser experience with (physical) health problems might explain their higher valuation of enjoyment of life.

Comparison with other research

In contrast to our study, most previous DCE-studies include the perspective of one stakeholder group, e.g., patients or healthcare workers, or compare the preferences of two stakeholder groups [22,23]. Furthermore, many health-related DCE-research include attributes related to characteristics of the new therapies or drugs (i.e., structure-attributes such as waiting time till appointment, care provider/setting or process-attributes such as shared decision-making) [24] whereas in the current study we included outcomes of the intervention.

In a previous paper, covering preference data from 8 European countries (including these Dutch data) [18], we also compared different stakeholder groups directly and reported considerable within-country agreement between stakeholders involved in integrated care with enjoyment of life ranking first and costs ranking last. However, we also found that patients assigned significantly higher values to physical functioning than professionals in five countries, which is in line with our finding that class 3, which focused more on physical health, contained the highest proportion of persons with multi-morbidity.

Similar to our study, other studies acknowledge the importance of measuring a broader set of outcomes than merely the physical and mental health outcomes that are traditionally included in health-related quality of life [25,26]. This is required to fully capture the outcomes that interventions are trying to achieve. The discussion on outcomes beyond the quality-adjusted life year (QALY) largely concentrates on interventions in the care sector, such as elderly care or care for physically or mentally disabled people. In that context, a lot of attention is being paid to well-being outcomes, for which several questionnaires were developed in recent years [27]. Well known instruments include the Adult Social Care Outcomes Toolkit (ASCOT) [28] and the ICEpop CAPability measure for Older people (ICECAP- O) [29,30]. The ASCOT focuses on social care related quality of life and, similar to our outcome measures, also includes ‘social participation’ as one of their 8 domains. The ICECAP-O is conceptually based on the capability approach and one of the five domains covered in this instrument is 'enjoyment' which received the highest weight in our study. A more recent instrument is the Well-being Of Older People measure [31] that captures relevant well-being domains for older people – among which multi-morbidity is common – and includes e.g., 'resilience and acceptance' and 'social contacts'. These outcomes were also included in the current DCE, in which resilience was in the top 3 outcomes that received the highest importance. Another example is the extension of the EQ-5D into the EuroQOL Health and Well-being (EQ-HWB) [32], which also includes outcomes in social care and carers' quality of life. Similarly, in our study we included social relationships and participation.

Strengths and limitations

This study is one of the first that elicited weights for a set of outcomes that goes beyond health, and requires trade-offs between health, well-being, experience, and costs to obtain weights. It included a sufficiently large representation of multiple stakeholder groups involved in integrated care for multi-morbidity. Furthermore, the inclusion of a variety of background characteristics and information on self-perceived health, allowed us to investigate the differences between stakeholders that had different opinions on the importance of the outcome measures. What is unique for this study is that we also asked the stakeholders for other roles they might have. We have learned that the additional perspective(s) that were chosen did not explain the variation in preferences between the classes.

Several limitations in the current study should also be mentioned. The survey could only be completed using a computer and not via a mobile phone. Therefore, younger persons may be underrepresented. Secondly, although the sample is quite large, payers are less well represented among the stakeholder groups. Moreover, although a DCE is a widely used method to elicit preferences in health care and health care delivery, it also has its limitations. One of the main concerns regarding DCEs is the external validity due to hypothetical bias, i.e., the disparity between stated preferences based on hypothetical DCE questions and revealed preferences based on actual choices in real life [33,34]. Recently, a number of case studies reported a high external validity of DCEs, with over 90% of the individual choices correctly predicted, thus suggesting a high degree of confidence [35]. However, this research does not pertain to integrated care, nor does it use outcome measures as attributes.

For future research, it would be interesting to further investigate the reasons behind differences in preferences. Preferences are, for example, likely to be influenced by a person's own experiences, or the experiences of significant others [36,37]. In the current study we did not explicitly ask about this.

Implications

Information about these preferences can be used in the design of new integrated care initiatives by concentrating on interventions that specifically aim to improve well-being and by better targeting interventions to patients' preferences. As this study has shown that patients' preferences cannot be presumed based on their characteristics, obtaining more insight in an individual's preferences should be an important part of a shared decision-making process.

The preferences can also be used in health care evaluation. Currently, health care evaluation focuses mainly on health status, life expectancy and QALYs, although there is a demand for tools that incorporate multiple outcomes that emerge from interventions with benefits beyond health [38,39]. The current study showed that outcomes related to well-being, and mental health in particular, were highly valued. More specifically, enjoyment of life received much weight in both the full sample analysis, and 3 out of 4 latent classes in which it received the highest or second highest weight. Yet it is not a common outcome measure in health care research. Future evaluations of integrated care interventions that measure a similar set of outcomes can make use of the weights obtained in this study. That enables the calculation of weighted outcomes which can be combined in an overall value score using multi-criteria decision analysis (MCDA) [11].

CONCLUSION

Stakeholders involved in integrated care for multi-morbidity value the outcome measure 'enjoyment of life' most and 'total health and social care costs' least. There is considerable heterogeneity in preferences, with a group of stakeholders assigning relatively higher importance to experience with care outcomes, a group assigning relatively higher importance to enjoyment of life and psychological wellbeing and a group focusing more on physical health. Differences in preferences were only weakly related to whether respondents were patients, informal caregivers, professionals, payers or policymakers as many stakeholders have multiple roles. This heterogeneity in preferences underlines the need to measure a wide range of different outcome measures when evaluating integrated care, including well-being outcomes and experience with care outcomes.

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APPENDIX

- **Appendix 1.** Latent class model

Outcome	Coefficient	Std. Err.	P-value	[95% Conf. Interval]	
Class 1					
Physical functioning level 2	1.201	0.288	0.000	0.636	1.766
Physical functioning level 3	2.065	0.435	0.000	1.213	2.918
Psychological well-being level 2	1.229	0.261	0.000	0.718	1.740
Psychological well-being level 3	2.737	0.423	0.000	1.909	3.566
Social relationships level 2	1.430	0.204	0.000	1.030	1.829
Social relationships level 3	2.202	0.281	0.000	1.652	2.753
Enjoyment of life level 2	2.165	0.252	0.000	1.672	2.659
Enjoyment of life level 3	3.511	0.410	0.000	2.706	4.315
Resilience level 2	2.165	0.252	0.000	1.670	2.660
Resilience level 3	3.031	0.311	0.000	2.422	3.641
Person-centeredness level 2	1.462	0.203	0.000	1.065	1.859
Person-centeredness level 3	2.689	0.289	0.000	2.123	3.255
Continuity of care level 2	2.149	0.237	0.000	1.684	2.613
Continuity of care level 3	2.777	0.275	0.000	2.237	3.317
Total costs level 2	0.394	0.160	0.014	0.080	0.709
Total costs level 3	1.208	0.222	0.000	0.773	1.643
Class 2					
Physical functioning level 2	1.380	0.129	0.000	1.126	1.633
Physical functioning level 3	1.926	0.174	0.000	1.584	2.268
Psychological well-being level 2	1.600	0.131	0.000	1.343	1.858
Psychological well-being level 3	3.264	0.190	0.000	2.892	3.636
Social relationships level 2	1.224	0.108	0.000	1.012	1.436
Social relationships level 3	1.830	0.128	0.000	1.578	2.081
Enjoyment of life level 2	3.061	0.192	0.000	2.684	3.438
Enjoyment of life level 3	4.778	0.267	0.000	4.254	5.301
Resilience level 2	1.480	0.118	0.000	1.248	1.712
Resilience level 3	2.028	0.149	0.000	1.737	2.319
Person-centeredness level 2	0.575	0.106	0.000	0.368	0.783

- **Appendix 1.** Continued.

Outcome	Coefficient	Std. Err.	P-value	[95% Conf. Interval]	
Person-centeredness level 3	0.848	0.128	0.000	0.597	1.099
Continuity of care level 2	0.961	0.112	0.000	0.741	1.180
Continuity of care level 3	1.185	0.130	0.000	0.931	1.439
Total costs level 2	0.469	0.091	0.000	0.290	0.649
Total costs level 3	0.835	0.119	0.000	0.602	1.069
Class 3					
Physical functioning level 2	2.571	0.225	0.000	2.130	3.012
Physical functioning level 3	3.819	0.316	0.000	3.199	4.440
Psychological well-being level 2	1.157	0.169	0.000	0.826	1.488
Psychological well-being level 3	2.583	0.249	0.000	2.095	3.070
Social relationships level 2	0.915	0.146	0.000	0.629	1.202
Social relationships level 3	0.836	0.189	0.000	0.465	1.207
Enjoyment of life level 2	1.757	0.188	0.000	1.389	2.126
Enjoyment of life level 3	2.722	0.264	0.000	2.204	3.240
Resilience level 2	1.552	0.184	0.000	1.192	1.912
Resilience level 3	2.110	0.227	0.000	1.665	2.556
Person-centeredness level 2	0.378	0.138	0.006	0.107	0.649
Person-centeredness level 3	0.595	0.175	0.001	0.251	0.939
Continuity of care level 2	0.558	0.156	0.000	0.251	0.864
Continuity of care level 3	0.819	0.174	0.000	0.478	1.160
Total costs level 2	0.332	0.139	0.017	0.059	0.605
Total costs level 3	0.654	0.177	0.000	0.306	1.001
Class 4					
Physical functioning level 2	0.150	0.102	0.139	-0.049	0.350
Physical functioning level 3	-0.022	0.119	0.856	-0.256	0.212
Psychological well-being level 2	0.142	0.093	0.127	-0.041	0.325
Psychological well-being level 3	-0.218	0.143	0.128	-0.499	0.063
Social relationships level 2	0.146	0.095	0.122	-0.039	0.332
Social relationships level 3	0.155	0.115	0.177	-0.070	0.380
Enjoyment of life level 2	0.504	0.105	0.000	0.298	0.711

- **Appendix 1.** Continued.

Outcome	Coefficient	Std. Err.	P-value	[95% Conf. Interval]	
Enjoyment of life level 3	0.485	0.140	0.001	0.210	0.760
Resilience level 2	0.558	0.098	0.000	0.366	0.750
Resilience level 3	0.615	0.117	0.000	0.385	0.845
Person-centeredness level 2	0.484	0.092	0.000	0.303	0.665
Person-centeredness level 3	0.756	0.123	0.000	0.515	0.997
Continuity of care level 2	0.936	0.114	0.000	0.713	1.158
Continuity of care level 3	1.321	0.129	0.000	1.068	1.573
Total costs level 2	0.318	0.087	0.000	0.148	0.487
Total costs level 3	0.641	0.102	0.000	0.441	0.842
share1					
_cons	-0.225	0.220	0.307	-0.657	0.207
share2					
_cons	0.428	0.183	0.019	0.070	0.787
share3					
_cons	-0.257	0.214	0.230	-0.677	0.163

“Without data, you’re just another person with an opinion.”

W. Edwards Deming

PART 2

Evaluation of a complex integrated care intervention for frail elderly

Chapter

4

Integrated Care For Frail Elderly: A Qualitative Study Of A Promising Approach In The Netherlands

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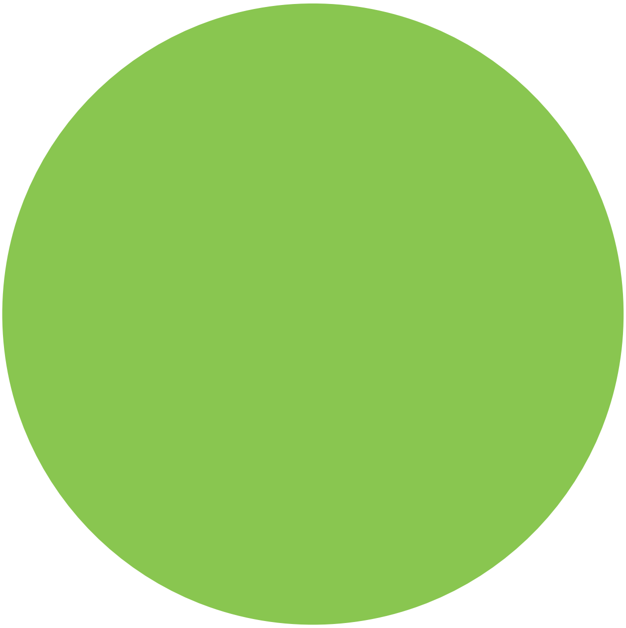

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ABSTRACT

Introduction: Increasingly, frail elderly need to live at home for longer, relying on support from informal caregivers and community-based health- and social care professionals. To align care and avoid fragmentation, integrated care programmes are arising. A promising example of such a programme is the Care Chain Frail Elderly (CCFE) in the Netherlands, which supports elderly with case and care complexity living at home with the best possible health and quality of life. The goal of the current study was to gain a deeper understanding of this programme and how it was successfully put into practice in order to contribute to the evidence-base surrounding complex integrated care programmes for persons with multi-morbidity.

Methods: Document analyses and semi-structured interviews with stakeholders were used to create a 'thick description' that provides insights into the programme.

Results: Through case finding, the CCFE-programme targets the frailest primary care population. The person-centred care approach is reflected by the presence of frail elderly at multidisciplinary team meetings. The innovative way of financing by bundling payments of multiple providers is one of the main facilitators for the success of this programme. Other critical success factors are the holistic assessment of unmet health and social care needs, strong leadership by the care groups, close collaboration with the healthcare insurer, a shared ICT-system and continuous improvements.

Conclusion: The CCFE is an exemplary initiative to integrate care for the frailest elderly living at home. Its innovative components and critical success factors are likely to be transferable to other settings when providers can take on similar roles and work closely with payers who provide integrated funding.

INTRODUCTION

Like many countries with a rapidly ageing population, the long-term care sector in the Netherlands is transitioning to improve efficiency and maintain affordability [1][2]. During the major reform in 2015 the long-term care sector was decentralised. The number of nursing homes was reduced considerably and access to nursing homes was restricted to those in need of 24-hour care. Municipalities became responsible for the provision of domestic home care and social support, whereas healthcare insurers became responsible for nursing care at home [3]. The reform stimulates elderly who were previously eligible for residential care and admission to nursing homes to stay at home longer, signalling the increased focus on self-sufficiency in our society. At the same time, the home care sector faced significant budget cuts [3].

As a result of the reform, a greater proportion of (frail) elderly is living at home with the support of primary care, home care, and informal care [3][4]. This population increasingly has a combination of physical, social and mental health problems [5]. Although, ageing in their own homes is generally in line with the preferences of elderly people, it also creates challenges [5-8]. The greater involvement of the municipalities in the funding of domestic and social care requires good communication and coordination between health and other care providers in order to prevent fragmentation or duplication that may lead to inefficient and ineffective care [9][10]. The collaboration between these providers is hampered by the traditional segmentation and 'silo-thinking' that is embedded in all aspects of the system [11][12]. There is no single professional or organisation that is truly responsible for coordinating care and support. Although GPs often take on this role, they do not always have a sufficient overview of, and time to explore, all available support services [13]. The increased complexity of frail elderly still living at home increases the number of visits to emergency departments and unplanned hospital admission and complicates the transfer of care when people return home [13][14]. Furthermore, the reform increased reliance on informal caregivers and thus alongside the possible benefits (e.g., feeling good about supporting a loved one, giving more meaning to one's life) the burden on these individuals increases (e.g., health problems, social isolation, withdrawal from the workforce) [14-18]. These challenges highlight the importance of improving the coordination of care within and across sectors in order to ensure efficient and high-quality care.

With the long-term care reform in sight, care providers in the Netherlands developed integrated care programmes for frail elderly. The development of these care programmes was stimulated by the Ministry of Health, which commissioned the National Care for the Elderly Programme that provided project-grants, and by healthcare insurers who offered additional funding for innovations [19-28]. The rise of these care programmes is accompanied by the need to evaluate such programmes, as healthcare insurers require evidence on their effectiveness in order to move from temporary to structural funding. However, these programmes are complex interventions and difficult to evaluate: they consist of multiple interacting components target multiple levels (individuals, groups, organisations, and systems), have a variety of intended outcomes that are largely impacted by the behaviour of those delivering and receiving the interventions, and are continuously adapted and improved [29-31]. Moreover, they often involve some form of budget pooling to break down the silos within and between health and social care. Previous studies have shown that elements from integrated care programmes are not always appropriately or fully implemented, or they worked out differently when put into practice [32-34].

The current paper provides an analysis of a thick description of a promising integrated care programme, the Care Chain Frail Elderly (CCFE), which is being implemented in the Netherlands. The CCFE is one of 17 innovative integrated care programmes being investigated in the EU-funded Horizon2020 SELFIE project (see **Box 1**). SELFIE aims to stimulate evidence-based implementation of integrated care for persons with multi-morbidity. The CCFE was selected because most frail elderly have multi-morbidity (i.e., co-occurrence of two or more chronic health conditions within one individual). The CCFE particularly met our selection criteria of being innovative in actively involving the individuals with multi-morbidity, their informal caregivers and the social care sector [41], which is what many integrated care initiatives are striving for. Sharing our in-depth understanding of the CCFE acquired through qualitative research may help to achieve this.

The purpose of this paper is to highlight the innovative elements of the CCFE and the factors that contribute to its success. It also aims to create awareness of the challenges involved in the implementation of the CCFE and how to address them. This leads to important insights that may inform future efforts to develop similar programmes in different settings and design evaluation studies.

Box 1

SELFIE was a Horizon2020 funded EU project that aimed to contribute to the improvement of person-centred care for persons with multi-morbidity by proposing evidence-based, economically sustainable, integrated care programmes that stimulate cooperation across health and social care and are supported by appropriate financing and payment schemes. More specifically, SELFIE aimed to:

- Develop a taxonomy of promising integrated care programmes for persons with multi-morbidity;
- Provide evidence-based advice on matching financing/payment schemes with adequate incentives to implement integrated care;
- Provide empirical evidence of the impact of promising integrated care on a wide range of outcomes using Multi-Criteria Decision Analysis;
- Develop implementation and change strategies tailored to different care settings and contexts in Europe, especially Central and Eastern Europe.

Seventeen promising integrated care programmes for persons with multi-morbidity are being evaluated in SELFIE using MCDA and a common set of core outcomes as well as programme-type specific outcomes. The latter depend on whether a programme is i) a population health management programme, ii) a programme targeting frail elderly, iii) a programme targeting persons with problems in multiple life domains, or iv) an oncology or palliative care programme.

The SELFIE consortium included eight organisations in the following countries: the Netherlands (coordinator) (NL), Austria (AT), Croatia (HR), Germany (DE), Hungary (HU), Norway (NO), Spain (ES), and the United Kingdom (UK). (www.SELFIE2020.eu) [Grant Agreement No 634288]

METHODS

Study design

In this study we qualitatively described a single case study applying a thick description: a qualitative empirical research method to investigate implicit social practices, such as care delivery, in their specific contexts [35]. A thick description covers several depths of analyses. The starting point is a formal description of the 'hard facts' based on document analyses. These written documents are often not sufficient to give a deeper understanding of what actually constitutes the programme below its surface when put into practice, i.e., the 'soft facts' on the 'how' and 'why'. For this purpose semi-structured interviews with key stakeholders are conducted. The interviews also complement the hard facts gathered in the course of the document analyses. When writing this manuscript we adhered to the COnsolidated criteria for REporting Qualitative research (COREQ) [36].

Procedure & data collection

The thick description method used in this study was centrally developed by the Austrian partner in the SELFIE consortium. During a SELFIE-meeting, they trained each partner-country in conducting thick descriptions. Specifically, interviewers were trained in using interview protocols and analysing the results. The one-day training focused on identifying relevant stakeholders, compiling interview protocols, and different methods of qualitative content analysis.

We studied a variety of documents about the care programme: official documents and contractual documents related to the programme, documents related to past evaluations, presentations given by project leaders, factsheets about the care programme and the collaboration between the care groups, a business case, documents regarding the bundled payment and other financial agreements, and documents about specific working groups related to the care programme. Most documents were provided by the project leader of the CCFE, others were publicly accessible on the internet.

For the interviews, we invited a purposive sample of 13 stakeholders via e-mail and/or phone. Two persons refused due to time constraints. Hence, over a 3-month period (July–September 2016), 11 semi-structured, face-to-face interviews were conducted with initiators of the care programme (n=2), programme managers (n=3), representatives of the payer organisations (n=2), medical and social care staff (n=2), an informal caregiver and a patient. An overview of the stakeholders and their reference is given in **Appendix A.1**. Interviews with professionals took place at their workplace, with the informal caregiver and patient they took place at their home. Interviews took between 33 and 62 minutes (mean 49 minutes). Five interviews were conducted by the first author (MH) and six by the first author together with a co-author (FL). No other persons were present during the interviews besides the interviewee and interviewer(s). These interviewers had a minimum of a Master's degree and experience in patient-contact and qualitative research. Prior to the interviews, authors had no established relationships with the interviewees; only with the programme managers there had been prior contact in order to prepare the participation of the CCFE in SELFIE and to identify stakeholders to interview.

For the different types of stakeholder groups, thematic focus areas were pre-defined across all SELFIE thick descriptions (see **Appendix A.2**), and a set of protocols for semi-structured interviews was prepared by the Austrian team and adapted to country/ programme specific issues by the Dutch team of SELFIE. By interviewing different types of stakeholders, we could gain insights into the programme from various perspectives. Interviewees were sent a topic list prior to the interview. Before the start of the interview, the interviewer(s) briefly introduced themselves and the SELFIE project. When new themes arose during the interviews, these were used in the interviews to come. The interviews were audio-recorded and transcribed verbatim. The transcripts were not returned to participants for correction, but the interviewees were sent the thick description and the quotes we used in the thick description.

Data analysis

All information retrieved from the document analysis was structured according to the conceptual framework for integrated care for multi-morbidity that was developed at the beginning of the SELFIE project, see **Appendix A.3** [37]. In the core of the framework is the holistic understanding of the person with multi-morbidity. This is surrounded by six components to systematically describe a care programme: service delivery, leadership & governance, workforce, financing, technologies & medical products, and information & research. The first author analysed the transcripts and discussed findings with two co-authors (FL and MRvM). Analysis was done using Mayring's content analysis method [38]. The transcripts were coded using mostly deductive coding as the topics were largely determined a priori. For each of the components of the framework, and for each topic described within the framework, sentences and paragraphs were selected that supplemented or illustrated the existing text. When new topics came up during the interviews, these were also coded and transformed into constructs. The first and second author separately coded the transcripts. The first draft of the thick description report was sent to the Austrian partner (TC, MK) and to the last author to provide feedback on the findings.

The thick description of the CCFE can be found on the SELFIE website (www.SELFIE2020.eu). The analysis presented in this manuscript focuses on the most innovative elements of the CCFE that characterise the programme.

Ethics statement

The Medical Ethical Committee of the Erasmus Medical Centre Rotterdam declared that this research was exempt from the Medical Research Involving Human Subjects Act. Participation was voluntary and could be retracted at any point. All participants signed an informed consent form, which was developed on the basis of the WHO informed consent for qualitative research and consisted of the following information: brief description of the SELFIE project, purpose and type of the research, participant selection, voluntary participation, procedure, duration, potential risks, benefits, reimbursements, confidentiality, sharing of the results, right to refuse/withdraw, and contact information.

RESULTS

The CCFE targets community-dwelling frail elderly with complex care needs.
Frailty is defined as a loss of functional abilities and control over one's life due to case and care complexity, which requires multidisciplinary care and case management. Subsequently, case complexity is defined as having complicated diseases, disabilities and frailty – often occurring simultaneously and difficult to diagnose. Care complexity refers to complicated care, for example due to a combination of needed care and no informal caregiver being present.

An overview of the programme and its components can be found in **Figure 1**. The CCFE programme started as a pilot in 2011 in a selected group of general practitioners. From 2013 onwards a wider implementation took place. The general goal of the frail elderly care programme is to provide person-centred care coordination and case management to keep frail elderly at home for as long as possible. An additional aim, formulated from the payers' perspective, is to develop structured multidisciplinary primary care that decreases the demand for secondary care, postpones nursing home admissions, and reduces health care costs for persons in this stage of life.

Below, the most noteworthy aspects of the CCFE are described per component of the conceptual framework.

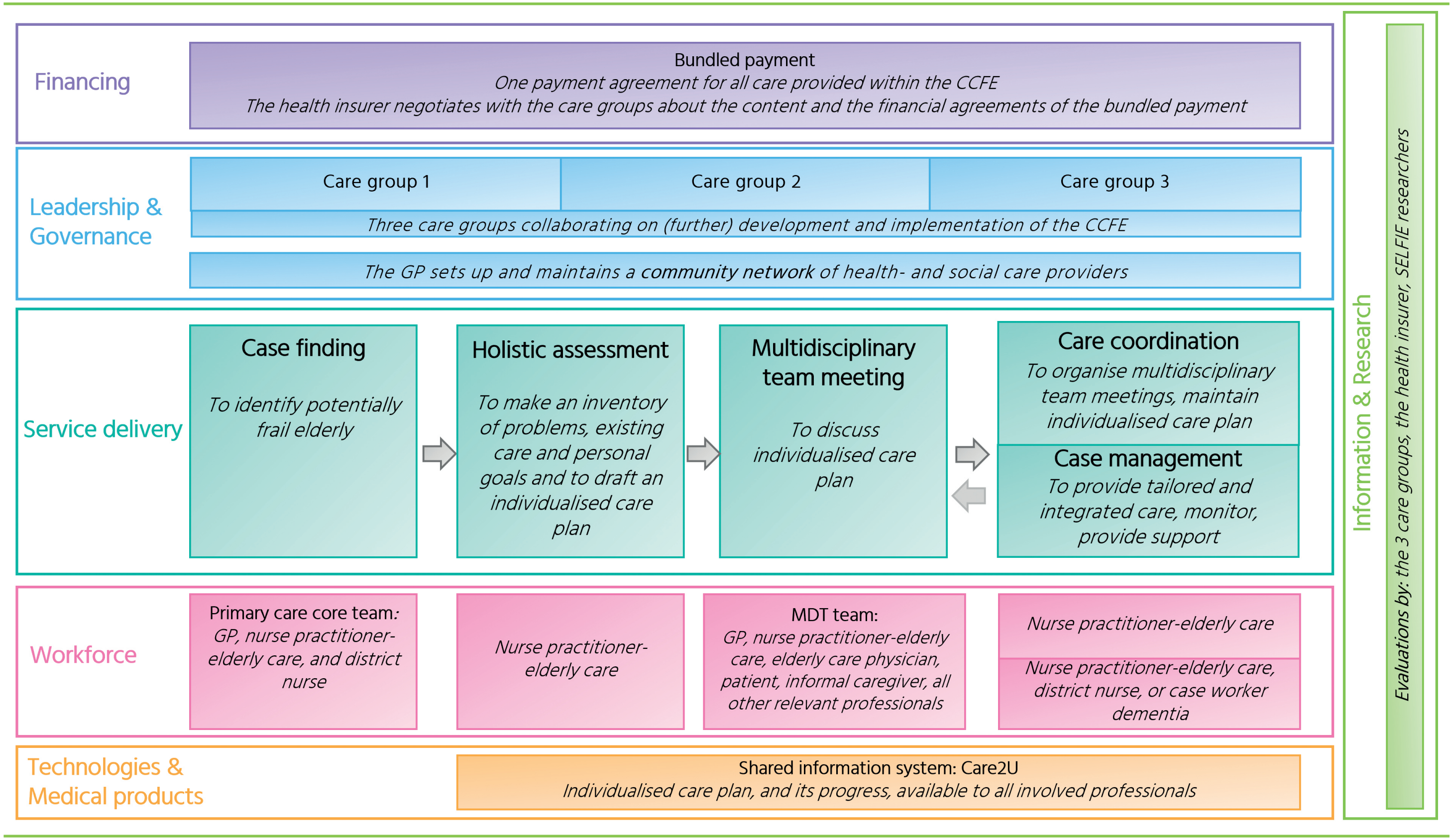
Service delivery

The CCFE is organised into four phases: (1) proactive case finding, (2) holistic assessment, (3) multidisciplinary team meeting(s), and (4) care coordination/case management. Three key aspects are discussed below.

Case finding

Potentially frail elderly persons are identified by a primary care core team of professionals using a case finding approach with inclusion criteria as defined above in a non-quantitative way. This approach includes a home visit to get a good overview of the health and social care needs. There is an agreement with the health care insurer that only the top 1% frail elderly of a GP-practice will be included in the programme. The insurer does not require a specific diagnosis for reimbursement, and instead trusts the professionals to select the elderly that will benefit most from the care programme.

Figure 1. Care process Care Chain Frail Elderly



Note: MDT = Multidisciplinary team

An advantage of this approach is that professionals are granted a lot of responsibility for including the 'right' patients. The central role of the primary care core team stimulates multidisciplinary collaboration between the GP-practice and home care. However, the 'soft' and un-protocoled method of selecting frail elderly can lead to issues; variations in the inclusion criteria across professionals have been observed, which is at the expense of uniformity. This variation partly results from the geriatric tool that is used to perform a holistic assessment of somatic, psychological, social, and communicative life-domains, as well as general functioning and self-sufficiency. Some professionals use this tool as a secondary inclusion criterion, whereas other professionals use the tool merely for guidance. One professional gave an example of an older person scoring high on the tool, but not being frail because the patient was handling the situation well. This professional argued that scoring is one thing, but the conversation between the professional and elderly is another:

> *"The instrument provides an indication of a situation, not necessarily an indication of problems. We should avoid being paternalistic in our approach." (PM_1)*

Thus, although the current case finding approach is likely to identify patients most in need of support, it may also increase practice variation.

Multidisciplinary team meetings

During the multidisciplinary team (MDT) meetings the individual care plan is discussed so that all professionals are aware of the elderly's goals, and the types of care/support the frail elderly person receives. This aims to ensure that all care providers are working towards the same goal in a proactive manner. Examples of interventions include: consultations with e.g., physiotherapist or dietician, weekly visits to a day care centre for elderly, arranging that a volunteer can help with e.g., groceries, medication review(s) by a pharmacist, or informal caregiver support.

The MDT meetings distinguish the CCFE from similar care programmes, because the frail older person and his/her informal caregiver are also invited to participate. Initiators, professionals and the patient and informal caregiver stress the importance of the involvement of the patient in the MDT-meeting for several reasons (*MS_2, IC_1, PM_1, MS_1, and FE_1*). The frail older person mentioned that he appreciates it and sees that all the professionals are collaborating with one-another. If anything was incorrect, he could "*jump in, and everyone listened to you.*" (*FE_1*) The informal caregiver stated that as a result of the multidisciplinary meeting:

> *"people at least communicated with one-another and looked at what was [...] really necessary for us".* (IC_1)

They further felt that they could be involved in the goal setting process. The patient and informal caregiver, and several other stakeholders, highlight the importance of their presence at the meeting to ensure a high quality of care and patient-centeredness and -involvement.

Downsides of their involvement, however, are that for patients it could be overwhelming to attend a meeting with all their caregivers at once, that the professionals need to adjust their professional language to the level of the patient (which could make the meeting take longer), that scheduling issues arise when multiple busy professionals and the patient and informal caregiver need to be present, and that MDT-meetings can only be used to discuss one case at a time. Furthermore, the healthcare insurer argues that if the patient is in good contact with his/her case manager, this should be sufficient. One of the initiators, who is also a care provider, stated that the added value of the patient's presence at the MDT-meeting remains inconclusive:

> *"Whenever the patient wishes to participate in the multidisciplinary team meeting, it will always be useful. However, it is not necessary for the patient to hear about technical details."* (IN_2)

Although more evidence on the added benefit of the patient's involvement in the MDT may be needed, it is hard to isolate the effect of this single component of the CCFE.

Care coordination and case management

The care programme separates two main care tasks. Care coordination supports the patient and his/her informal caregiver in keeping an overview and navigating through the system. As most of the frail elderly's care is complex, multiple professionals are involved, which enhances the care burden for that patient. Case management monitors the execution of the individual care plan, signals additional needs, and further adapts the care to the patient's wishes and needs.

Both tasks are usually carried out by the same professional, the nurse practitioner specialised in elderly care, which is beneficial for the continuity of care. This is not a requirement and there is a degree of flexibility. In some cases, the district nurse is appointed as case manager, usually because she was already involved in the care process and is the caregiver who visits the elderly most frequently. An advantage of having this professional take on this role, is that collaboration between the GP-practice and home care organisations, where the district nurse is employed, is stimulated. Despite this, initial evaluations revealed difficulties in the collaboration between the district nurse and the GP-practice. One of the difficulties relates to data-sharing. District nurses need to record data in in the shared information system 'Care2U', as well as in their own information system, causing duplicate registration. Another difficulty is that GPs have to work with several district nurses in their community, and the turnover of district nurses is rather high. Thus, GPs have to maintain many collaborations and give many district nurses access to their agendas. As time passed, close physical proximity to the GPs and the electronic medical records appeared to result in fewer district nurses fulfilling the tasks of case manager.

Leadership & governance

Key elements of leadership & governance are the role of care groups and the community networks (see **Figure 1**).

Care groups

Unique to the CCFE, and perhaps more generally to Dutch primary care provision, is the role of care groups. A care group is a group of primary care providers that cooperate in the provision of chronic care and support GPs in implementing care pathways. Healthcare insurers contract a care group, and not the individual GPs. Care groups either employ or subcontract professionals who provide the care.

Three care groups in the Netherlands developed the CCFE. This governance has given the care groups a strong position to negotiate with the healthcare insurer about the content, price and quality of care. Alongside the benefits in relation to cross-disciplinary collaboration and financing, the collaboration between the three care groups also enhances the uniformity of frail elderly care within the region, because they aligned their ideas surrounding frail elderly care. Using a uniform approach is important, especially since many professionals are involved:

> *"A district nurse has to deal with several GP-practices and the GP-practices have to deal with several home care organisations and different teams. To make it even more complicated, there are just a lot of parties. That is why we decided to work in a uniform way. (IN_1)*

Although uniformity is achieved to a large extent, differences between the care groups and between the types of GP-practices remain. One example is the difference in organisational culture and leadership. The fact that care groups either employ or subcontract the professionals results in different scopes of influence a care group has on the care being provided. Furthermore, some GPs are running a practice in a small village, whereas others work in a large group practice in a big city, affecting the efforts required to create community networks.

Community network

As the target population consists of frail elderly living at home, a great amount of cross-sector collaboration from formal and informal care providers is required. So far, professionals in the network had primarily been working alongside one-another without actually collaborating. The reforms of the long-term care sector in the Netherlands tried to stimulate collaboration between health and social care. The CCFE has embraced this trend and required that GPs set-up community networks. Professionals recognise the importance of a close collaboration between sectors. One of the district nurses describes the benefits:

> *"The good thing about our collaboration is that we know each other very well. When I'm with a client and I notice something special, I can call the GP and I know that I will be heard, because we know each-other."* (MS_1)

The ease and effectiveness of collaboration between GP-practices and the social care sector, specifically with district nurses, differs between GP-practices. A professional responsible for setting up a community network mentions that it may be easier to work in a small town, in a smaller setting, because it is easy to identify possible partners in the care-chain to collaborate with (MS_2). Also, for some GP-practices, the collaboration with home care organisations was already established before the start of the CCFE, making it easier to reach out to these organisations (MS_1). As a result of implementing the CCFE, this collaboration became more structural with meetings being held on a regular basis (MS_1). However, the collaboration with the welfare organisations, which for example provide community-based volunteer-support, is not yet optimal for reasons related to privacy protection when sharing information and the large amount of these organisations. Nevertheless, the importance of establishing community networks is widely recognised.

Workforce

In the CCFE a differentiation is made between the primary care core team and a wider network of professionals that can be called upon in the multidisciplinary care team.

New multidisciplinary teams of professionals

The *primary care core team* consists of the GP, nurse practitioner and district nurse, and meets once or twice a month to discuss potentially frail elderly. The role of this team is to signal frailty within- and outside of the healthcare sector and match care accordingly.

The *multidisciplinary care team* discusses the individualised care plan based on the personal goals of the elderly. Possible professionals involved are the nurse practitioner, GP, district nurse, elderly care physician, physical therapist, psychologist, case worker dementia, pharmacist, speech therapist, occupational therapist, and/or geriatrician. Whomever is already involved in the care process, is invited to attend the MDT-meetings, including the older person and his/her informal caregiver. This ensures person-centred, integrated, and coordinated care as all care providers agree on the same care plan.

Key professionals

The nurse practitioner is one of the key professionals in the core team. This nurse has followed an educational programme to specialise in elderly care. In the CCFE she is involved in each step of the care process, works in close collaboration with the GP, and also maintains contact with other professionals involved in the care process. For most frail elderly she is the main contact point. Tasks appointed to the nurse practitioner in the CCFE are: case management, care coordination, setting up a community network (in collaboration with the GP), and process monitoring of transfer care, polypharmacy, and data collection of quality indicators on the patient level. She has a certain amount of hours to spend on elderly care, next to her other tasks as nurse practitioner.

The elderly care physician plays an important advisory role in the multidisciplinary care team. This is a relatively new medical professional working in primary care in the Netherlands. (S)he is specialised in frail elderly care and has experience working in multidisciplinary teams and with advance care planning. The elderly care physician has an important role in the programme in coaching the GP and nurse practitioner and acting as a source of information for them. She reviews the results of the holistic assessment and the individualised care plan and is available for home visits and consultations with other professionals. The rationale behind involving these professionals in the CCFE is that they will improve the knowledge and skills of the GP and nurse practitioner, which will gradually make their own role smaller over time.

The structural embedding of both care teams into the CCFE and the important role of the key professionals has greatly stimulated cross-sector collaboration and they are seen as valuable assets.

Role of the informal caregiver

At the national level there is a trend towards a greater role for the informal caregiver; the CCFE aims to unburden the informal caregiver by recognising the potential burden and ensuring adequate support to prevent drop-out of the informal caregiver and hospitalisation of the frail elderly. One of the goals of support is to ensure that the positive aspects of informal caregiving (satisfaction) outweigh the burden. According to one of the professionals, elderly are not always aware of the severity of their frailty and the amount of support they need from their informal caregiver(s) (MS_2). The role of the nurse practitioner is then to convince the frail elderly to accept formal care:

> *"We are trying to meet the needs of the patient, yet also to unburden the informal caregiver and increase the safety of the patient."* (MS_2)

Financing

To incentivise integration of care, the programme is funded via a bundled payment contract that each care group negotiates separately with the healthcare insurer. The bundled payment is a fixed annual budget that should cover all frail elderly care. It is based on three factors: an average tariff per frail elderly based on the estimated number of minutes of care, agreed upon between insurer and care group (care group specific, confidential), overhead costs (care group specific, confidential), and an estimated number of frail elderly included in the programme (care group specific). It is agreed upfront that this number should not exceed 1% of the GP-practice. The bundled payment covers care provided by the GP, nurse practitioner, pharmacist (for medication review), geriatrician (consultation by phone), and the physician assistant. It also includes tasks not directly related to a patient, e.g., setting up the community network. It does not include care provided by the district nurse, elderly care physician, case manager dementia, physiotherapist, occupational therapist, social worker and welfare worker. These professionals are funded in the usual way, either by the healthcare insurers (Health Insurance Act) or by the municipality (Social Support Act).

An important facilitator to the implementation of the CCFE were the macro-level reforms that supported this exploration of new ways of financing elderly care. The bundled payment is a great improvement in financing, both compared to the fee-for-service payment for consultations in the past and to the short-term project-based way of financing integrated elderly care. It is seen as an innovative and sustainable way of financing integrated care for community-dwelling frail elderly. The advantage of the bundled payment from the perspective of an insurer is that it allows them to contract the care group, and care programme, as a whole, instead of contracting all individual GPs and care activities separately (HI_2). Not only does this result in a lower administration burden, the insurer can also delegate the monitoring of the care delivered by the GP to the care group. The insurer believes it is easier for a care group to steer and monitor a GP, since a care group is managed by care providers that easily relate to the GPs (HI_2). Furthermore, the bundled payment results in predictable costs for the insurer because the size of the target population is predictable and the costs of the bundled care are known. However, the bundled payment contract has to be renewed annually and the burdensome negotiations about what care to include and for which tariffs start over again. Nevertheless, the key elements of the care programme have not changed much since the beginning:

> *"In 2010/11 we invented the care programme, and at this moment, the key elements are the same. [...] We still think it is best to include the patient in the multidisciplinary team meeting, and the goals in the individualised care plan are the patient's goals and the professional's... These elements remain always a topic of discussion [with the insurers], but we keep coming back to the same quality requirements." (PM_1)*

Both the care groups and the insurer do have plans about the further development of the CCFE. The insurer encourages the care groups to further differentiate the reimbursement for GPs, based on the case mix of patients in their practice. The care groups do not see a fundamental reason to do so, as it goes against the basic idea of integrating payments and it would increase the difficulties in administration (*PM_3*). The care groups would rather see that all activities not directly related to the care programme - for example treatment of ear syringing - are reimbursed outside the bundled payment. The healthcare insurer does not want to reimburse all consultations separately, because it is hard to define the boundaries of frail elderly care.

The influence of the insurer has been a challenge, but is also an asset as it provides promise for the financial sustainability of the programme. A debated point between the insurer and care groups is the continuous request of convincing effectiveness evidence. Furthermore, the presence of the frail elderly and informal caregivers at the MDT-meetings is being debated (*HI_1, HI_2*) (see also *Service delivery*). In contrast to the professionals who are generally very positive towards their presence, the healthcare insurer has pointed out the lack of evidence on the benefits, making it difficult to secure funding for this element of the programme (*HI_2*). After speaking with frail elderly and informal caregivers in other regions where they are not involved in the MDT meetings, the insurer did not get the impression that these elderly were any less satisfied and these programmes were less costly (*HI_1*).

Finally, it has to be acknowledged, that although the bundled payment incentivises collaboration between professionals, the scope of the current bundle is limited. It only includes reimbursement for a few professionals, mostly operating in primary care. In the future it would be desirable to expand this.

Information & research

The healthcare insurer has emphasised that evaluation plays an indispensable role in the future of the bundled payment and discussions with the care groups (HI_1). Very preliminary findings from a qualitative study by the insurer suggested that there is room for improvement in the way the ICT is organised and the collaboration with social care. Overall, the direction of the results was quite positive, which was beneficial for the continuation of the care programme.

The care groups have also tried to evaluate the programme. They can extract quality indicators from the shared information system, but also collected data from a small sample of patients. However, these evaluations had limitations concerning the number of respondents, lack of control group, and the small scope of outcomes. Although all parties are aware of the importance of evaluating the care programme, they also recognise the difficulties, e.g., selection bias, measuring patient-reported outcomes (PROMs) and experience (PREMs) in frail elderly, and spill-over effects making it difficult to identify an appropriate control group.

Technologies & medical products

In the CCFE the information system 'Care2U' is used, that offers a secured platform to share information between the professionals involved. There is a direct link between the GP information systems and Care2U, which saves a lot of time. Professionals not working at the GP-practice, however, have to log their proceedings in their own information system separately. Also, a district nurse working with GPs from the three different care groups, has three accounts to log into Care2U. These difficulties are seen as challenges:

> *"It is possible for the various chain partners to use Care2U, but to make it work on an organisational level, there should be financial incentives. For example, that registration for reimbursement is done in Care2U." (PM_1)*

Each individual care plan is posted in Care2U and is accessible for all involved professionals. The frail older person needs to approve that professionals can access Care2U. Professionals have different degrees of access; some professionals only need to have access to specific information, such as a dietician, whereas other professionals need to have access to all information. Care2U enables the nurse practitioner to monitor appointments and tasks of partners in the care chain, for example to see if these professionals have met certain deadlines (e.g., for lab results).

The frail elderly can make use of a patient portal that was developed to support self-management. It is not yet possible to link this patient portal to Care2U, and thus does not provide access to the individualised care plan. One of the care groups has started a pilot to create this link and offers elderly the opportunity to report their experience in the individual care plan directly (PM_1).

Having a shared information system for professionals and a patient portal are two potentially influential facilitators for the CCFE to succeed. Nevertheless, if these systems create extra work, they may function as a barrier. Thus, ensuring that professionals can easily use these and that they save time will make the programme attractive and efficient.

DISCUSSION

In this study we systematically described and analysed a promising integrated care approach for frail elderly along the lines of the six components of the SELFIE framework for integrated care for persons with multi-morbidity. The CCFE programme has several factors in common with other integrated care programmes for frail elderly, e.g., a holistic assessment, individualised care plan, multidisciplinary care, care coordination, and/or case management [19-28], but is innovative and distinguishes itself in that it targets the *frailest* GP-population, invites the frail elderly and their informal caregivers to participate in the MDT-meeting, and is funded through a new bundled payment system.

Challenges and facilitators

One of the challenges of the care programme during implementation was the harmonisation of the case finding process across care groups in order to ensure inclusion of a similar population of frail elderly. Relying on professional judgement may be an efficient way to ensure inclusion of the frail elderly most in need of better support and most responsive to change. In the current approach, elderly are included when they are already very frail. In the future it might be desirable to identify elderly with an increased risk of becoming frail earlier on, in order to focus more on prevention and create better long-term results [39].

Part of the case complexity of the frailest elderly is related to their social environment. Therefore, the development of a community network requires further attention. The care programme offers time and funding for this task. District nurses seem to be well equipped for this because they have a good overview of services offered and collaborating with other parties in the community is part of their everyday work [40]. However, GPs still struggle with initiating and maintaining collaborations with the social care providers because they do not have much experience with this.

The presence of the patient and informal caregiver at the MDT-meeting is the ultimate expression of person-centred care and crucial for shared decision-making. Although it is an essential element of the care programme, it complicates planning of the meetings and increases their duration and costs, and could therefore be a barrier to sustainable implementation.

The CCFE has the means to address these challenges. Namely, a dedicated staff and management, the bundled payment and the Care2U ICT-system. Extending the bundled payment to include a wider variety of services is possible, especially for healthcare services covered by the healthcare insurers. The inclusion of social care services is more difficult as it would require breaking down the funding silos between health and social care, the latter of which is covered by the municipality. The Care2U ICT-system facilitates cross-sector collaboration because all professionals involved in the care provision have access to it, with different disciplines having access at different levels. Compatibility with ICT-systems outside the care groups is a hurdle to overcome. Its further development is funded by the bundled payment.

Strengths and limitations of the study

This in-depth qualitative analysis of the CCFE programme, incorporated the perspectives from multiple stakeholders, including professionals, managers, payers, a patient and an informal caregiver. This contributed to a broad insight into the evolution of the programme in daily practice, which commonly deviates from the plans on paper. We used a purposeful sample of interviewees and we did not continue recruiting respondents until data saturation was reached. However, we did get a broad overview of different views although it is possible that more critical views are less well represented. Nevertheless, it is not the aim in qualitative research to attain a representative sample.

Furthermore, our analysis encapsulates a moment in time in the continuous adaptation and improvement of the programme. Examples of adaptations currently being implemented are optimisation of transfer care to and from hospitals, more frequent medication reviews, better integration of dementia care, and further development of a patient portal to give the elderly access to their individual care plan. Sharing the lessons learned at this point in time may help others to better tailor their own programme to their context.

Future evaluation

Although the CCFE is considered to be a programme with great potential, more quantitative evidence is needed to secure its sustainability. Therefore, we have designed a prospective quasi-experimental study comparing the programme to usual care on the Triple Aim. This study is a Multi-Criteria Decision Analysis in which patient-reported outcomes are not only measured, but also weighted by their importance to different stakeholders [41].

CONCLUSION

This study presents essential success factors of implementing an integrated care programme for community-dwelling frail elderly, the CCFE. These success factors include the holistic assessment of unmet health and social care needs, direct engagement of the patient in the multidisciplinary team meetings, strong leadership by the care groups, close collaboration with the healthcare insurer, a bundled payment, a shared ICT-system and a shared desire to continuously improve. The extent to which these factors are transferable to other settings depends on the context. However, our general recommendations for implementing a similar intervention in different health and social care systems are to adopt an incremental growth approach, involve a GP-role that is responsible for building a team culture and maintaining close relationships with both the patient and the social care sector, establish an integrated way of financing that secures budgets for a longer term, and design a shared information system to accommodate a smooth collaboration between all professionals involved in the programme.

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APPENDIX

- **A.1:** Interview partner overview

Reference in text	Stakeholder description
PM_1	**P**rogramme **m**anager/ Initiator
PM_2	**P**rogramme **m**anager
PM_3	**P**rogramme **m**anager
IN_1	**In**itiator
IN_2	**In**itiator/ Other stakeholder
HI_1	Representative of the sponsor/payer (**H**ealth **i**nsurer)
HI_2	Representative of the sponsor/payer (**H**ealth **i**nsurer)
MS_1	Non-physician **m**edical **s**taff
MS_2	Non-physician **m**edical **s**taff
IC_1	**I**nformal **c**aregiver
FE_1	Patient (**F**rail **E**lderly)
Refused participation due to time constraints	Physician
Refused participation due to time constraints	Representative of the sponsor/payer (Health insurer)

A.2: Thematic focus areas for interviews

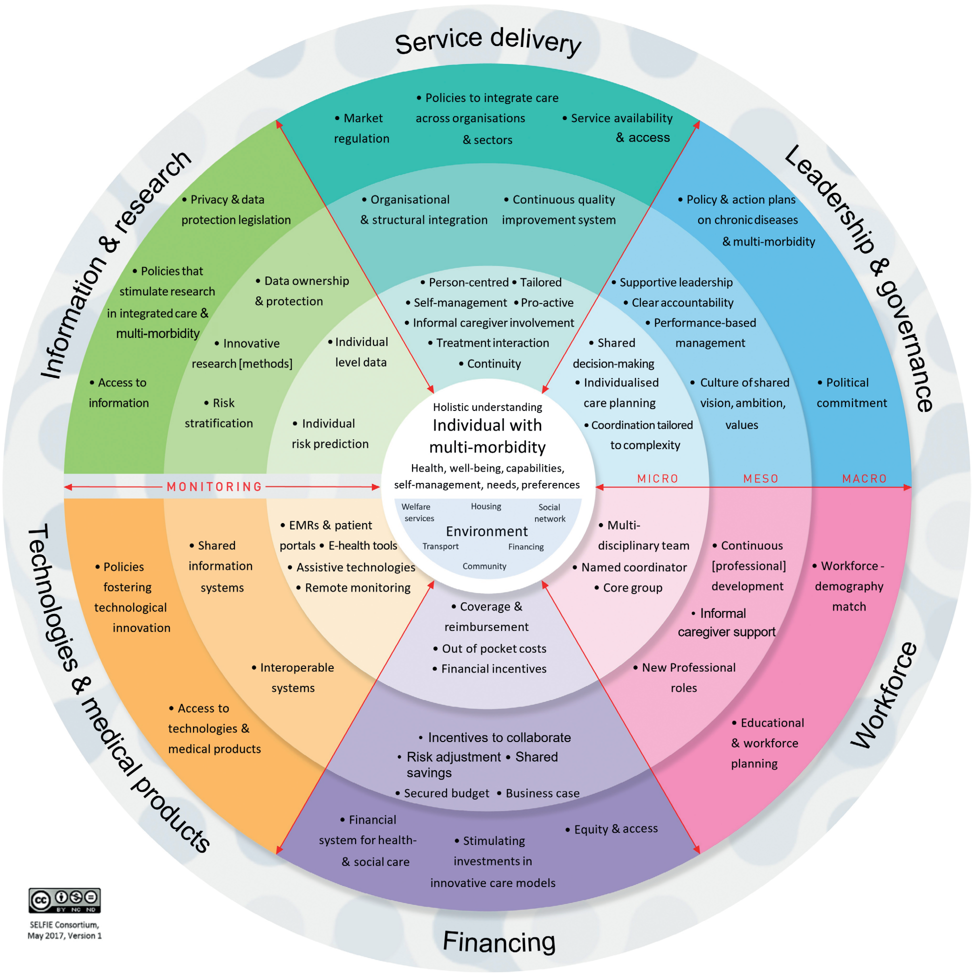
Stakeholder \ Task		T2	T3	T4	T5	T6	T7	T8*	TS**
		Implementation process and barriers to implementation	Design of delivery of care including relationships between partners involved	Partnerships beyond the health care system	Use of ICT applications	Use of self-management interventions	Involvement of new professional roles	Evaluation	Financing and payment schemes
A.	Manager(s)	X	XX	XX	X	XX	XX	X	X
B.	Initiator(s)	XX	XX	X	o	X	XX	X	X
C.	Representative(s) of sponsor/payer organisations	XX	o	X	o	o	o	X	XX
D1.	Physician(s)	o	XX	XX	X	X	X	X	X
D2.	Non-physician medical staff (e.g. nurses), social staff, new professional groups (if applicable)	o	XX	XX	XX	XX	XX	X	X
E.	Informal caregiver(s)	o	XX	XX	XX	XX	o	o	o
F.	Client(s) or their representative(s)	o	XX	XX	XX	XX	XX	o	o
G.	(Other stakeholder(s))	*Interview protocol to be developed only if necessary and based on protocols of the stakeholder types closest to this stakeholder*							

Note: XX = main question; X = to be asked only if not mentioned in the answers to main questions; o = not necessary to be asked

* results of T8 mostly derived from document analysis; in the interviews, we want to know about the experience with evaluation/monitoring

** results of TS partly derived from document and literature analysis

● **A.3:** The SELFIE Framework for Integrated Care for Multi-Morbidity



SELFIE Consortium,
May 2017, Version 1

Chapter 5

Value-Based Person-Centred Integrated Care For Frail Elderly Living At Home: A Quasi-Experimental Evaluation Using Multi-Criteria Decision Analysis

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ABSTRACT

Objective: To evaluate the value of the person-centred, integrated care programme Care Chain Frail Elderly (CCFE) compared with usual care, using Multi-Criteria Decision Analysis (MCDA).

Design: In a 12-month quasi-experimental study, Triple-Aim outcomes were measured at 0, 6 and 12 months by trained interviewers during home-visits.

Setting: Primary care, community-based elderly care.

Participants: 384 community-dwelling frail elderly were enrolled. The 12-month completion rate was 70% in both groups. Propensity score matching was used to balance age, gender, marital status, living situation, education, smoking status, and three-month costs prior to baseline between the two groups.

Intervention: The CCFE is an integrated care programme with unique features like the presence of the elderly and informal caregiver at the multidisciplinary team meetings, and a bundled payment.

Primary and secondary outcomes measures: The MCDA results in weighted overall value scores that combines the performance on physical functioning, psychological well-being, social relationships and participation, enjoyment of life, resilience, person-centeredness, continuity of care, and costs, with importance weights of patients, informal caregivers, professionals, payers, and policymakers.

Results: At 6 months, the overall value scores of CCFE were higher in all stakeholder groups, driven by enjoyment of life (standardised performance scores 0.729 versus 0.685) and person-centeredness (0.749 versus 0.663). At 12 months, the overall value scores in both groups were similar from a patient's perspective, slightly higher for CCFE from an informal caregiver's and professional's perspective, and lower for CCFE from a payer's and policymaker's perspective. The latter was driven by a worse performance on physical functioning (0.682 versus 0.731) and higher costs (€22,816 versus €20,680).

Conclusions: The MCDA indicated that the CCFE is the preferred way of delivering care to frail elderly at 6 months. However, at 12 months, MCDA-results showed little difference from the perspective of patients, informal caregivers, and professionals, while payers and policy makers seemed to prefer usual care.

INTRODUCTION

The Care Chain Frail Elderly (CCFE) is an integrated care programme for community-dwelling frail elderly in the Netherlands that offers person-centred care, coordination, and case management to support elderly in living at home for as long as possible.[1] It was developed as part of a movement towards decentralisation of long-term care, increased self-sufficiency, and societal participation that was stimulated by the Dutch government to maintain affordability of elderly care. This movement led to a reform of the long-term care sector in 2015.[2] The number of nursing- and residential homes was reduced considerably and access to nursing homes was restricted to those in need of 24-hour care. Municipalities became responsible for the provision of homecare and social support, and health insurers for financing nursing care at home. While the importance of homecare was growing, this sector was confronted with significant budget cuts.[3] The reform accelerated the development of integrated-care initiatives, which were spearheaded by the National Care for the Elderly-Programme commissioned by the Ministry of Health.[4,5]

Integrated care for older people often comprises similar components, namely, comprehensive assessment, individualised care planning, case management, and multidisciplinary team meetings.[6,7] All of these elements are also present in the CCFE. In addition, the intervention is unique in several respects. First, the older person and their informal caregiver are present at the multidisciplinary team meetings where the results of the comprehensive assessment and the individual care plan are discussed. Second, the CCFE is financed by a bundled payment aiming to stimulate collaboration between professionals. The bundled payment is a fixed amount of money per patient that covers all services provided by the GP, nurse-practitioner and physician assistant, regardless of diagnoses, medication review by the pharmacist, telephone consultation by the geriatrician, non-individual-patient-related activities such as building a community network, and overhead. Third, it targets the top-1% frailest elderly registered in a GP-practice who are living at home with complex care needs, using a case-finding approach. The CCFE aims to better integrate care across sectors and build a network of support around the patient, and thereby to improve their physical, mental, social health and well-being and experience with care. Ideally, this also reduces secondary care and residential long-term care utilisation and thereby costs. The CCFE has previously been described in detail elsewhere.[1]

Although such integrated-care programmes are designed to meet the older person’s needs, previous studies show mixed (cost-)effectiveness results.[8–10] A potential explanation is that common methods to assess effectiveness tend to focus on measuring traditional outcomes, such as physical functioning, whereas that is not the primary focus of these programmes.[8,11] Cost-effectiveness analyses measure Quality-Adjusted Life-Year (QALY) gains, which is also less appropriate because integrated care for frail elderly focuses more on well-being than on survival and health-related quality of life.[12] For example, elderly are stimulated to visit outpatient day-care activities to enhance their social participation, or their experience with care is improved by individual care planning and helping them navigate through the health- and social-care system. Accordingly, empirical evaluations should include these domains to accurately value the potential benefits of an intervention.[13,14]

Therefore, we adopted a broader evaluation method, namely Multi-Criteria Decision Analysis (MCDA) to evaluate the CCFE. MCDA is innovative in elderly care because it synthesises a wide variety of outcome measures, in this case patient-reported health and well-being measures (PROMs), experience with care measures (PREMs), and costs. Together, they cover the Triple Aim.[15] In the MCDA outcomes are weighted by their importance according to five stakeholder groups: patients, informal caregivers, professionals, policymakers, and payers. The weighted performance scores are aggregated into overall value scores.[16]

METHODS

Study population

The study population consisted of community-dwelling frail elderly, where frailty is defined as being in need of complex care due to loss of functional abilities and control over one's life. To participate in the intervention, they had to be registered at a GP-practice from one of three care groups (i.e., an association of primary care providers that cooperate in the provision of chronic care) that offer the CCFE. They had to be able to comprehend study information and answer questions, either independently or with the help of an informal caregiver or trained interviewer.

Intervention

A primary care team consisting of the GP, nurse-practitioner, and district nurse, identifies potential candidates for the CCFE using a case-finding approach. To be included in the CCFE, the health insurer does not require a specific diagnosis or the use of a screening tool. They trust that the GP and nurse practitioner know their patients and are most suited to select the elderly that may benefit from the care programme. Furthermore, there is an agreement with the health insurer that only the top 1% frail elderly of a GP-practice will be included in the programme. Subsequently, the nurse-practitioner visits the older person at home and performs a comprehensive geriatric assessment of the needs, capabilities, and preferences in the physical, psychological, cognitive, and social domains. Depending on this assessment, the nurse-practitioner consults professionals in the community (e.g., physical therapists, occupational therapists, social workers, elderly care physicians, geriatricians, dementia case workers) and arranges informal care support. Together with the frail elderly and informal caregiver, the nurse-practitioner drafts an individual care plan, largely driven by the elderly's personal goals.

The nurse-practitioner organises multidisciplinary team meetings to discuss the individual care plan with all professionals involved. The frail elderly and their informal caregiver participate in these meetings. During the first meeting a case manager is assigned, a role mostly taken up by the nurse-practitioner. The case manager is the main contact point, monitors the execution of the individual care plan, and further adapts the care to the patient's wishes and additional needs. Once enrolled, an elderly person usually stays in the CCFE until nursing home admission or if they pass away.

To support collaboration between professionals, the CCFE uses 'Care2U', a secured ICT-platform to share information. Professionals have different degrees of access and the elderly must approve access. Care2U includes the individual care plan and is used by the nurse-practitioner to monitor appointments and services of providers in the care chain.

The CCFE is financed by a bundled payment contract between each care group and the dominant health insurer in the region and is re-negotiated every year. These care groups are legal entities of primary care providers who develop chronic care programmes, support the provision of these programmes and are contracted by the health insurer to coordinate chronic care in a region.

Study design

The MCDA was conducted as part of a 12-month prospective quasi-experimental study comparing two parallel groups:

Intervention group included frail elderly enrolled into the CCFE between April 2017-August 2018 and were recruited to participate in the study by either their GP or nurse-practitioner.

Control group consisted of frail elderly receiving usual care, recruited at GP-practices from one of three participating care groups. These practices had not (yet) implemented the CCFE. To ensure a similar level of frailty in both groups, the GP-practices in the control group applied the same case-finding approach. They were assisted by a GP specialised in elderly care that had experience with the CCFE.

Self-reported outcome and experience measures and care utilisation data were gathered at baseline and after 6 months and 12 months, during face-to-face interviews at the elderly's home, performed by trained interviewers. The interviewers were not involved in delivering any aspects of the intervention but were aware of which group the respondent belonged to. Data collection took place between April 2017-August 2019.

Outcome measures

Table 1 gives an overview of all outcome measures. These outcomes were selected based on a literature review, workshops with representatives from the five stakeholder groups, and focus groups with individuals with multi-morbidity, and measured with validated questionnaires.[17]

Costs

Health- and social-care utilisation data were obtained with the iMTA-Medical Consumption Questionnaire, which includes questions about contacts with GP, nurse-practitioner, GP-assistants, physiotherapists and other paramedical therapists, dieticians, psychologists, social workers, welfare workers, and medical specialists, hospital admissions, rehabilitation, homecare, residential care and nursing homes, and informal care during the past three months.[26] Unit costs were largely based on reference prices from the Dutch Costing Manual.[29] Medication costs were based on prescription data from GP-information systems, which were combined with unit costs from Dutch drug database '*G-Standaard*', using ATC-codes.[30] Programme costs of the CCFE were based on the bundled payment contracts between each care group and the dominant health insurer.[1] Elderly from the control group could participate in single-disease care programmes for diabetes, cardiovascular risk management or Chronic Obstructive Pulmonary Disease. The proportion of elderly participating in these programmes was obtained from Care2U and average prices of the bundled payments were obtained from the Dutch health claims database (Vektis).[31]

Statistical analysis

Comparability of groups

We applied inverse probability weighting to increase the comparability of the intervention and control groups at baseline.[32] The logistic regression model to estimate the propensity score included age, gender, marital status, living situation, educational level, smoking status, and costs three months prior to baseline as a proxy for complexity. The propensity score p reflects the estimated probability of an individual to be in the intervention group. By setting the weight for individuals in the intervention group to 1, and for individuals in the control group to $p/(1-p)$, we estimated the average treatment effect in the treated (ATT) in the weighted mixed effect models described below.[33] To assess the comparability of the two groups, we calculated the mean percentage standardised bias, the Rubin's B (absolute standardised difference of the means of the linear index of the propensity score in the intervention group and matched controls), and the Rubin's R (ratio of intervention group and matched control group variances of the propensity score index). For sufficient balance, B should be less than 25% and R be between 0.5 and 2.[34]

Table 1. Outcome measures and instruments

Core set^	**Instrument to measure outcome**	**Scale**
Health & Well-being		
Physical functioning	Activities of Daily Living (ADL) (Katz-15)[18]	0-15 (worst)
Psychological well-being	Mental Health Inventory (MHI-5)[19]	0-100 (best)
Enjoyment of life	Investigating Choice Experiments for the Preferences of Older People (ICECAP-O)[20]	1-4 (best)
Social relationships & participation	Impact on Participation & Autonomy (IPA), social life and relationships domain[21]	0-28 (worst)
Resilience	Brief Resilience Scale (BRS)[22]	6-30 (best)
Experience of care		
Person-centeredness	Person-centered Coordinated Care Experience Questionnaire (P3CEQ), experience of person-centered care domain[23]	0-18 (best)
Continuity of care	Nijmegen Continuity Questionnaire (NCQ), team and cross boundary continuity domain[24] + Client Perceptions of Coordination Questionnaire (CPCQ)[25]	1-5 (best)
Costs		
Health, social, and informal care costs	iMTA Medical Consumption Questionnaire[26]	
Medication costs	Prescriptions in patient records extracted from GP information systems	
Bundled payments and chronic care programmes	Care chain information system 'Care2U'	
Additional outcomes		
Autonomy	Pearlin Mastery Scale[27]	7-35 (best)
Burden of medication	Living with Medicines Questionnaire - LMQ[28]	0-10 (worst)

^The core set of outcomes was measured across all studies included in the SELFIE project. For these outcomes, weights were elicited, and these outcomes were included in the MCDA. The additional outcomes were not included in the MCDA.

Treatment effects

Treatment effects were estimated using weighted mixed effect models with a random intercept at individual level (as we had longitudinal data) and the following covariates: time, intervention, an interaction term for time and intervention, age, gender, marital status, living situation, educational level, and smoking status. This combination of matching and regression adjustment has been shown to best reduce covariate imbalance between groups.[35] The mixed effects models were used to predict the mean scores of the outcome measures in both groups at each time point, assuming the control group had the same baseline score as the intervention group to directly compare both groups. All statistical analyses were performed using STATA16.1.

MCDA

In the MCDA, the predicted mean scores of the outcomes at 6- and 12-months follow-up were standardised on a 0-1 scale to remove differences in measurement scales, using relative standardisation, see **Appendix 1**. For all outcomes in the MCDA, a higher score indicates better performance. The standardised outcomes were weighted by their importance and subsequently summed to obtain an overall value score for the intervention and control group separately. The relative importance-weights were elicited in an online weight elicitation study among patients, informal caregivers, professionals, payers, and policymakers, using a discrete choice experiment (DCE).[36] The relative weights of the outcome measures included in the MCDA by stakeholder group can be found in **Appendix 2**.

Sensitivity analysis

To assess the joint uncertainty in outcome scores and importance-weights, we performed a probabilistic sensitivity analysis using Monte Carlo simulation. We used Cholesky decomposition to obtain 10,000 replications of both the standardised outcomes and the importance-weights. From this, we obtained the 95%-confidence intervals around the overall value scores for each stakeholder group. Additionally, we calculated the proportion of MCDA-iterations in which the CCFE has a higher overall value score than usual care.

Patient and Public Involvement

Patients were involved in the selection of outcome measures, which was largely based on focus groups as described above.[17] In multiple National Stakeholder Workshops held during the entire process of the study, stakeholders from the five stakeholder groups were asked to reflect upon the outcome measures, the study design and data collection, and the results of the study. The study design was set-up in close collaboration with care providers to ensure feasibility of the data collection. Before the start of data collection, the questionnaire was piloted in a frail older person. The authors disseminated results via conference presentations. Results of this study were also disseminated to participating care providers, patients and informal caregivers using communication methods other than scientific papers, i.e., by e-mail and newsletters disseminated at the GP-practices.

Ethics approval and consent to participate

The study was conducted as one of the case studies in Horizon2020-project SELFIE (Sustainable intEgrated care modeLs for multi-morbidity: delivery, FInancing and performancE).[37] The study protocol was reviewed by the medical ethics committee of the Erasmus Medical Centre, the Netherlands. The committee concluded that the rules laid down in the Dutch Medical Research Involving Human Subjects Act do not apply to this research (*MEC-2017-121*). All study participants have provided informed written consent.

RESULTS

Respondents

Figure 1 presents the flow chart of patients included in the study. The retention rate at 12-month follow-up was 70% in both groups. Main reasons of loss-to-follow-up were also similar in both groups and included death, burden of study participation, and cognitive incapacity.

- **Figure 1.** Flowchart patient participation

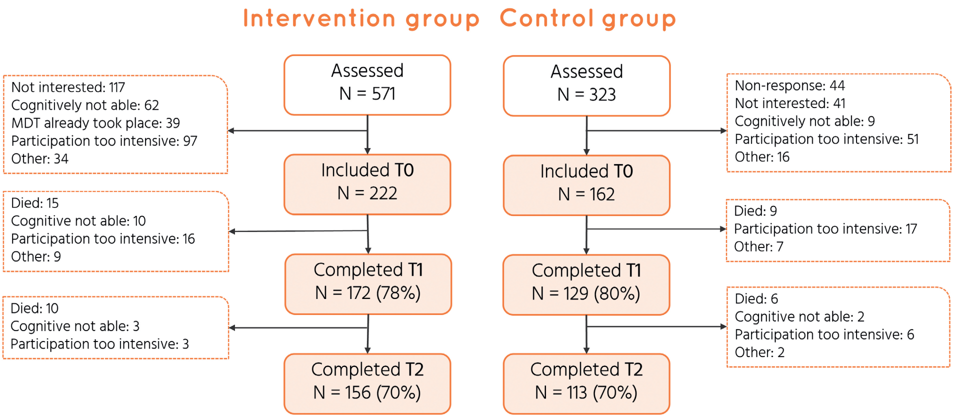

Table 2 presents the baseline characteristics of respondents before and after inverse probability weighting. After IPW, the matching statistics were within the desired range (Rubin's B < 25%, Rubin's R 0.5 – 2). No substantial differences between the groups

remained. The common support graph shows a good overlap in propensity scores, see **Appendix 3**.

Table 2. Baseline characteristics before and after inversed probability weighting

	CCFE N=222	**UC N=162**	**St. diff.**	**UC N=162**	**St. diff.**
Baseline characteristics	**Before IPW**			**After IPW**	
Age, mean	83.4	84.8	-0.224	83.6	-0.028
Female (%)	63.5	64.2	-0.014	64.8	-0.027
Marital status (%)	3.6	3.7	-0.005	2.6	0.054
• Single, never married	44.6	40.7	0.078	45.0	-0.008
• Married or living together	44.6	50.6	-0.120	47.1	-0.049
• Widow(er)	7.2	4.9	0.095	5.4	0.078
• Divorced					
Living situation (%)	54.1	61.1	-0.143	55.2	-0.024
• Independent, alone	46.0	38.9	0.143	44.8	0.024
• With others					
Educational level (%)	70.3	70.4	-0.002	73.3	-0.067
• Low	20.3	14.8	0.143	14.3	0.158
• Medium	9.5	14.8	-0.164	12.4	-0.090
• High					
Current smoker (%)	14.4	8.6	0.181	13.8	0.019
Outcome measures at baseline, mean	**Before IPW**			**After IPW**	
Physical functioning (0-15) ^	4.4	4.5	-0.032	4.5	-0.052
Psychological well-being (0-100)	71.4	71.7	-0.018	70.6	0.040
Enjoyment of life (1-4)	2.8	2.9	-0.137	2.9	-0.143
Social relationships and participation (0-28) ^	9.2	8.0	0.345	8.2	0.280
Resilience (6-30)	19.3	19.0	0.062	19.0	0.082
Autonomy (7-35)	22.3	22.1	0.054	21.9	0.098
Burden of medication (0-10) ^	2.1	2.4	-0.111	2.5	-0.166
Person-centeredness (0-18)	11.7	12.5	-0.209	12.7	-0.246
Continuity of care (1-5)	3.7	3.8	-0.140	3.8	-0.182
Total costs 3 months prior to the study (€) ^	5,453	5,267	0.028	5,631	-0.026

Table 2. Continued.

	CCFE N=222	UC N=162	St. diff.	UC N=162	St. diff.
Statistics to assess matching#	**Before IPW**			**After IPW**	
Mean bias			11.9		5.1
Rubin's B			39.7		21.4
Rubin's R			1.06		1.10

^ Higher score indicating worse performance; St. diff = Absolute Standardised Mean Difference, also called Absolute Standardised Bias; # On variables used in PSM, i.e., age, gender, marital status, living situation, educational level, smoking, total costs 3 months prior to the study. CCFE=Care Chain Frail Elderly. UC=Usual care. IPW=Inverse Probability Weighting

Treatment effects

Table 3 reports the estimated treatment effects of the CCFE at 6- and 12-months of follow-up. Results show that the CCFE improved person-centeredness at both time points. At 6 months follow-up, physical functioning declined in both groups but even further in the intervention group. At 12 months follow-up, the CCFE performed worse on autonomy and burden of medication. The predicted mean performance scores of all outcomes on their natural scale can be found in **Appendix 4**.

Costs

Table 4 presents details on the mean costs after inversed probability weighting at 6- and 12-months, from a healthcare and a societal perspective. From a healthcare perspective, after 6 months, costs were €751 higher in the intervention group than in the control group. After 12 months, the costs were €1,796 higher. These differences were largely due to programme costs and costs of homecare.

When adopting a societal perspective, 6-month costs were €662 higher in the intervention group than in the control group, whereas 12-month costs were €2,136 higher. Costs of informal care were slightly lower in the intervention group than in the control group after 6 months, but higher after 12 months.

● **Table 3.** Treatment effects at 6- and 12-months follow-up

	6 months			**12 months**		
Core set of outcomes included in the MCDA	**Estimated change CCFE**	**Estimated Change UC**	**Diff in change Mean (95% CI)#**	**Estimated change CCFE**	**Estimated change UC**	**Diff in change Mean (95% CI)#**
Health & Well-being						
Physical functioning (0-15) ^	0.74	0.27	0.47 (0.06 ; 0.88)*	1.33	0.95	0.39 (-0.10 ; 0.87)
Psychological well-being (0-100)	-0.64	-0.34	-0.30 (-3.86 ; 3.25)	-1.35	-0.10	-1.26 (-5.20 ; 2.68)
Enjoyment of life (1-4)	0.10	-0.08	0.18 (-0.02 ; 0.38)	0.05	0.02	0.04 (-0.18 ; 0.25)
Social relationships & participation (0-28) ^	0.11	0.41	-0.31 (-1.04 ; 0.43)	0.26	0.61	-0.36 (-1.25 ; 0.54)
Resilience (6-30)	-0.02	-0.21	0.19 (-0.63 ; 1.01)	0.03	0.11	-0.08 (-1.01 ; 0.85)
Experience of care						
Person-centeredness (0-18)	1.10	-0.38	1.48 (0.57 ; 2.38)*	1.33	0.00	1.33 (0.18 ; 2.49)*
Continuity of care (1-5)	0.10	-0.02	0.12 (-0.05 ; 0.28)	0.16	0.03	0.13 (-0.05 ; 0.31)
Costs						
Total health and social care costs^	5,405	4,745	660 (-1,650 ; 2,970)	17,223	15,206	2,017 (-2,361 ; 6,395)
Additional frail elderly-specific outcomes	**Estimated change CCFE**	**Estimated change UC**	**Diff in change Mean (95% CI)#**	**Estimated change CCFE**	**Estimated change UC**	**Diff in change Mean (95% CI)#**
Autonomy (7-35)	-0.50	0.02	-0.52 (-1.38 ; 0.34)	-0.49	0.55	-1.04 (-1.95 ; -0.14)*
Burden of medication (0-10) ^	0.13	-0.21	0.34 (-0.36 ; 1.04)	0.18	-0.61	0.78 (0.04 ; 1.53)*

*^ Higher score indicating worse performance; $^{\#}$ based on robust SE; * $p<0.05$. MCDA=Multi-Criteria Decision Analysis. CCFE=Care Chain Frail Elderly. UC=Usual care. CI=Confidence interval.*

Table 4. Mean costs (€) after 6- and 12-months of follow-up (after IPW)

	6 months			12 months		
Cost category	**Mean costs (SD) CCFE (n=172)**	**Mean costs (SD) UC (n=129)**	**Difference between means (SE)**	**Mean costs (SD) CCFE (n=156)**	**Mean costs (SD) UC (n=113)**	**Difference between means (SE)**
Chronic care programme(s) #	534 (-)	72 (-)	462 (-)	1,068 (-)	143 (-)	925 (-)
Homecare	3,289 (4,371)	3,158 (4,016)	131 (481)	7,597 (8474)	6,330 (7680)	1,267 (980)
Long-term care admissions	969 (5,411)	766 (4,980)	203 (589)	1,624 (7,959)	2,051 (10,025)	-427 (11,33)
Hospital admissions	670 (2,670)	485 (1,833)	185 (259)	1,199 (3,603)	1,119 (2,992)	80 (403)
Emergency room visits	112 (308)	116 (341)	-4 (38)	179 (372)	196 (418)	-17 (49)
Outpatient day-care activities	235 (1,180)	310 (1,481)	-74 (170)	627 (2,642)	408 (1,846)	218 (285)
Medical specialist care	338 (619)	317 (408)	21 (60)	631 (789)	609 (615)	23 (86)
Paramedical care (e.g., physiotherapist)	529 (636)	481 (548)	48 (72)	1,059 (1,113)	1,005 (1,133)	55 (145)
General practitioner ~	21 (27)	288 (272)	-267 (28)	25 (34)	492 (394)	-467 (44)
Medication	363 (662)	318 (551)	45 (65)	744 (1,232)	605 (927)	139 (124)
Subtotal costs from healthcare perspective	**7 060 (8,441)**	**6,310 (7,671)**	**751 (922)**	**14,753 (14,498)**	**12,957 (14,185)**	**1,796 (1,777)**
Informal care	3,690 (5,780)	3,779 (7,318)	-89 (891)	8,063 (12,436)	7,723 (13,643)	340 (1,783)
Total costs from societal perspective	**10,750 (10,381)**	**10,089 (11,003)**	**662 (1,309)**	**22,816 (19,050)**	**20,680 (20,251)**	**2,136 (2,551)**

 # Costs for chronic care program(s) is an average estimation which is the same for each respondent in their respective group; ~ Costs for GP-care in the intervention group are largely included in the costs of the chronic care programme. SD=Standard deviation. SE=Standard Error. CCFE=Care Chain Frail Elderly. UC=Usual care.

Multi-Criteria Decision Analysis

Table 5a and b present the standardised outcome scores for the CCFE and usual care as well as these scores weighted according to each stakeholder's importance-weights, at 6- and 12-months, respectively. The overall value scores show that all stakeholder groups preferred the CCFE over usual care at 6 months follow-up. This was driven by the performance scores of person-centeredness and enjoyment of life, and the high importance-weight of the latter. In more than 75% of iterations the CCFE had a higher overall value score than usual care. At 12 months follow-up the difference disappeared, and the probability that the CCFE had a higher overall value score dropped below 50% for payers and policymakers. This was driven by worse scores in physical functioning and costs.

DISCUSSION

Main findings

The MCDA has shown that all stakeholders preferred the CCFE over usual care at 6 months with a likelihood of having a higher overall value score of over 75%. This was mainly driven by higher performance on enjoyment of life and person-centeredness, and the great importance of the former outcome. Results became more diffuse at 12 months. Patients were indifferent, informal caregivers, and professionals slightly favoured the CCFE, whereas payers and policymakers demonstrated a slight preference for usual care. This was mainly due to a worse performance of the CCFE on physical functioning and costs. When looking at the disaggregated scores, person-centeredness was consistently higher in the CCFE. Physical functioning deteriorated in the intervention group at 6 months, but this effect disappeared at 12 months. When some outcomes improve whereas others deteriorate, the current MCDA approach is a suitable method to aggregate them into overall value scores that vary depending on the importance that stakeholders assign to the different outcome measures. These results show that the CCFE is the preferred way of delivering care to frail elderly if improvements in enjoyment of life and person-centeredness are considered more important than physical functioning and costs.

Context and comparison with previous evaluations

As this is the first MCDA of a frail elderly programme, it is impossible to directly compare the value scores to other studies. MCDA also provides insight in the disaggregated effects of the CCFE, but these are hard to compare to other studies as well, due to the very frail target group of the CCFE, the different contexts in which the interventions are implemented, the different intervention components, and outcome measures[38,39] Regarding the context of the CCFE, we should stress that the programme was implemented in a setting with a strong primary care sector. GPs already have a

- **Table 5a.** Value scores in the Multi-Criteria Decision Analysis at 6 months, using DCE weights

Outcome measures			**Patients**		**Partners**		**Professionals**		**Payers**		**Policymakers**	
	Standardised performance score#		**Weighted score**		**Weighted score**		**Weighted score**		**Weighted score**		**Weighted score**	
	CCFE	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**
Physical functioning	0.672	0.740	0.108	0.118	0.074	0.081	0.081	0.089	0.094	0.104	0.094	0.104
Psychological well-being	0.706	0.709	0.120	0.120	0.106	0.106	0.127	0.128	0.127	0.128	0.106	0.106
Enjoyment of life	0.729	0.685	0.168	0.157	0.182	0.171	0.160	0.151	0.175	0.164	0.160	0.151
Social relationships and participation	0.718	0.696	0.057	0.056	0.065	0.063	0.079	0.077	0.072	0.070	0.072	0.070
Resilience	0.711	0.704	0.107	0.106	0.099	0.099	0.092	0.091	0.078	0.077	0.099	0.099
Person-centeredness	0.749	0.663	0.060	0.053	0.060	0.053	0.060	0.053	0.045	0.040	0.060	0.053
Continuity of care	0.718	0.696	0.072	0.070	0.086	0.083	0.079	0.077	0.057	0.056	0.072	0.070
Costs (societal care perspective)	0.685	0.729	0.021	0.022	0.041	0.044	0.041	0.044	0.055	0.058	0.048	0.051
Overall value scores			**0.711**	**0.702**	**0.713**	**0.700**	**0.719**	**0.708**	**0.703**	**0.696**	**0.711**	**0.702**
95% Confidence interval^			0.702 - 0.721	0.692 - 0.712	0.703 - 0.723	0.690 - 0.710	0.702 - 0.722	0.691 - 0.711	0.699 - 0.721	0.692 - 0.714	0.700 - 0.721	0.692 - 0.712
% CCFE > UC^			82%		90%		87%		75%		81%	

● **Table 5b.** Value scores in the Multi-Criteria Decision Analysis at 12 months, using DCE weights

Outcome measures			**Patients**		**Partners**		**Professionals**		**Payers**		**Policymakers**	
	Standardised performance score#		**Weighted score**		**Weighted score**		**Weighted score**		**Weighted score**		**Weighted score**	
	CCFE	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**
Physical functioning	0.682	0.731	0.109	0.117	0.075	0.080	0.082	0.088	0.095	0.102	0.095	0.102
Psychological well-being	0.701	0.713	0.119	0.121	0.105	0.107	0.126	0.128	0.126	0.128	0.105	0.107
Enjoyment of life	0.711	0.703	0.164	0.162	0.178	0.176	0.157	0.155	0.171	0.169	0.157	0.155
Social relationships and participation	0.720	0.694	0.058	0.056	0.065	0.062	0.079	0.076	0.072	0.069	0.072	0.069
Resilience	0.706	0.709	0.106	0.106	0.099	0.099	0.092	0.092	0.078	0.078	0.099	0.099
Person-centeredness	0.744	0.668	0.060	0.053	0.060	0.053	0.060	0.053	0.045	0.040	0.060	0.053
Continuity of care	0.719	0.695	0.072	0.069	0.086	0.083	0.079	0.076	0.058	0.056	0.072	0.069
Costs (societal care perspective)	0.673	0.739	0.020	0.022	0.040	0.044	0.040	0.044	0.054	0.059	0.047	0.052
Overall value scores			**0.707**	**0.707**	**0.708**	**0.706**	**0.714**	**0.713**	**0.698**	**0.702**	**0.706**	**0.707**
95% Confidence interval^			0.696 - 0.717	0.697 - 0.717	0.697 - 0.718	0.695 - 0.716	0.697 - 0.717	0.695 - 0.716	0.695 - 0.717	0.697 - 0.719	0.695 - 0.717	0.696 - 0.718
% CCFE > UC^			50%		57%		54%		39%		48%	

^ based on Monte Carlo simulation; # colour scheme ranges from red (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care.

history of collaborating with other primary care providers, for example by working jointly in health centres. Hence, setting up community networks and collaborating in wider multidisciplinary teams was not such a big step. When implementing similar interventions in countries with a less strong primary care system, collaboration may require more efforts. On the other hand, the potential for savings due to the intervention might be higher in countries where the GPs does not act as a gatekeeper to secondary care, as a programme like the CCFE could substitute more secondary care services by primary care services.

Despite differences between studies, most previous studies did not find effects on physical functioning.[8,40] This may be expected as these programmes rarely aim to achieve improvements in this domain. As the CCFE aimed to improve experience with care, the sustained improvement in person-centeredness found in this study suggests that the programme has fulfilled that aim.[1] The worsening in autonomy and burden of medication seems counterintuitive. A possible explanation for the deterioration in autonomy is that as elderly in the CCFE were confronted with their frailty, e.g., by discussing their needs for support, they became more aware of their loss of control. This could have led to lower autonomy scores in the intervention group, especially due to the self-report method; measurement by a third party might have led to different scores.[41] The higher medication burden in the intervention group could be explained by alterations in medication after the medication review, which may have led to (temporary) side-effects.

One could question the relevance of finding a sustained effect on an outcome (i.e., person-centredness) that was less highly valued than other outcomes in the DCE. This may raise the question whether the aim of the CCFE was well-targeted. However, weights were derived from a DCE that asks respondents to choose between two hypothetical care programmes, which gives them the opportunity to trade-off person-centredness for, for example, improved physical functioning. Even though this is likely to be an appropriate reflection of their preferences if all options were open, in real life this trade-off may no longer exist, because improving physical functioning might not be possible anymore. Hence, there is a discrepancy between what is important to a patient and what is feasible in practice.

Strengths & limitations

One of the strengths of our study was its controlled study design. Defining an appropriate control group to evaluate ongoing programmes for frail elderly is a challenge.[8,42] In the current study, a potential limitation concerns the case finding approach to identify the target population of both groups, namely frail elderly in need of complex care due to loss of functional abilities and control over one's life.

In the intervention group this was done by GPs offering the programme and in the control group by GPs not offering the programme. To ensure a similar level of frailty in both groups, the latter GPs were assisted by a GP specialised in elderly care. This has been successful as the baseline characteristics of both groups were quite similar. It is especially important in evaluating frail elderly care, as there is commonly little room for actual improvements in health, and prevention or delay of deterioration can only be shown in comparison with a control group.

Another strength was the data collection on a broad range of PROMs and PREMs by interviewers who made a total of 954 home-visits, which was a major endeavour. Collecting patient-reported data did limit the generalisability of the results, as some frail elderly could not participate in the evaluation because that was too burdensome or impossible. Hence, the frailest among the elderly were not represented in this study, especially not those with dementia. The attrition rate in our study was relatively low, i.e., 30% across both groups at 12 months. We did observe that respondents in the control group that were lost to follow-up were slightly older and had worse physical functioning at baseline compared to non-dropouts in the control group (and overall), see **Appendix 5**. This may have led to an underestimation of the treatment effect.

A further strength of our study was the detailed cost-analysis, even including costs of medication, social care and informal care, which are often excluded from other studies.[8] This analysis showed a cost increase reflecting the greater investment of resources to support frail elderly in ageing in place which is of great importance to many older persons.

In the design and reporting on the MCDA we followed the good practice guidelines as laid out by the ISPOR MCDA-taskforce.[43] Strengths of MCDA are that it enables explicit, transparent and accountable decision-making, i.e., for every decision what was valued most and by whom can be tracked down as well as whether this was due to improvements in certain domains or a higher relative importance of a particular outcome. Furthermore, MCDA makes it possible to include additional elements of value that go beyond health or QALYs, which is especially important for complex interventions with multiple aims such as improving well-being and experience with care. However, the consequence is that we may favour interventions that achieve improvements in these outcomes above interventions that have greater health outcomes. This may be justifiable for elderly care. Such an argument would raise another point that is debated in MCDA, namely whether or not to include costs in the overall value score.[44,45] To elicit a weight for costs, stakeholders had to trade-off costs against other outcomes, which makes the relative contribution of costs to the overall value score explicit. However, it can be argued that this does not adequately address the opportunity costs of the CCFE.[46] We also performed the MCDA without costs as a sensitivity analysis, see **Appendix 6**, which led to higher overall value scores for the CCFE at both time points. However, now the overall value forms a composite benefit score for which a new cost-effectiveness threshold must be determined. Although we believe the current set of outcomes captures the full potential value of an integrated care programme for frail elderly, this set should be tailored to each intervention's aims and target group. Therefore, when the set of outcome measures changes, new thresholds need to be determined. Another option is to calculate the cost-per-value and prioritise interventions with the lowest cost-per-value ratio, but this only leads to a ranking of interventions.[45] A last point of discussion on MCDA is that it requires a deliberative component to avoid making decisions based solely on the model.[45] In our study we presented results from five stakeholder perspectives which inevitably calls for further deliberation to determine which perspective should prevail.

Implications

Although the CCFE does not improve the (physical) health of patients, it is still positively evaluated by all stakeholder groups at 6 months. At 12 months stakeholders were mainly indifferent. This warrants further research into interventions to maintain the effects of such programmes in the long-term. Furthermore, we advocate a wider use of MCDA to evaluate multi-faceted, person-centred, integrated care programmes for frail elderly that aim to improve multiple outcomes, including those that go beyond health. MCDA enables a transparent and explicit decision-making process and serves as a tool to help decision-makers reach a decision. Therefore, MCDA-results are a good starting point for deliberation before deciding upon reimbursement or broader implementation of new interventions.

CONCLUSION

After 6 months, the overall value score for the CCFE was higher than for usual care across all stakeholders, but at 12 months, the preference for the CCFE had disappeared. The CCFE led to sustained improvements in enjoyment of life and person-centeredness, which is aligned with the program's aim, but also to a deterioration in physical functioning at 6 months and higher costs. Therefore, the CCFE is only the preferred way of delivering care to frail elderly in case improvements in enjoyment of life and person-centeredness are considered more important than costs and physical functioning.

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SUPPLEMENTARY FILES

- **Appendix 1:** Equation relative standardisation

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

Where

x = predicted mean score on the natural scale

a = the intervention group

b = the control group

j = outcome measure *j*

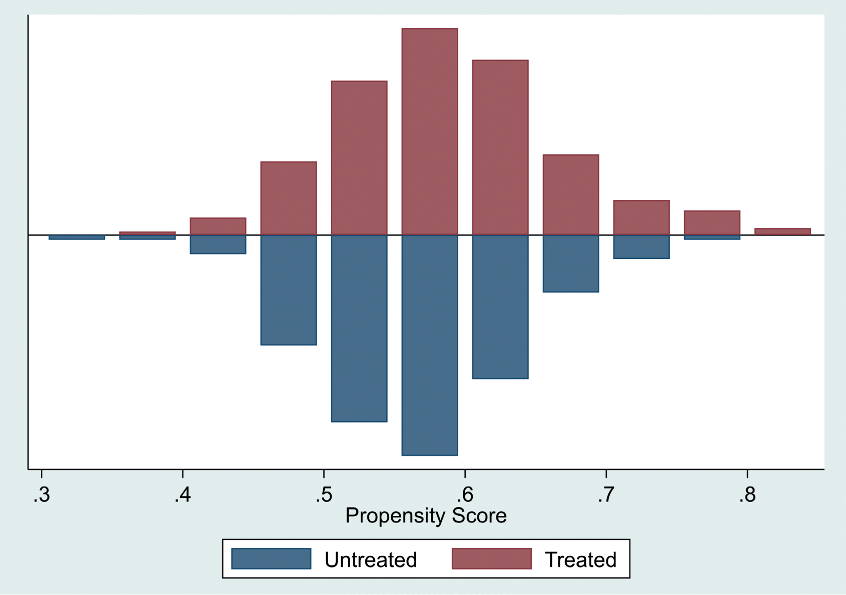
Note: For all outcomes in the MCDA, a higher score indicates better performance. To achieve this, in the above-mentioned equation, x is replaced by $1/x$ for outcomes where a higher score on the natural scale indicates a worse performance (e.g., costs).

- **Appendix 2:** Relative DCE weights (0-1) (SE) of the outcomes used in the MCDA by type of stakeholder

	Patients (n=156)	**Partners (n=158)**	**Professionals (n=155)**	**Payers (n=104)**	**Policymakers (n=151)**
Physical functioning	0.16 (0.15)	0.11 (0.12)	0.12 (0.12)	0.14 (0.13)	0.14 (0.14)
Psychological well-being	0.17 (0.15)	0.15 (0.14)	0.18 (0.16)	0.18 (0.17)	0.15 (0.15)
Enjoyment of life	0.23 (0.18)	0.25 (0.19)	0.22 (0.19)	0.24 (0.23)	0.22 (0.19)
Social participation & relationships	0.08 (0.10)	0.09 (0.11)	0.11 (0.11)	0.10 (0.11)	0.10 (0.10)
Resilience	0.15 (0.13)	0.14 (0.13)	0.13 (0.12)	0.11 (0.10)	0.14 (0.13)
Person-centeredness	0.08 (0.10)	0.08 (0.10)	0.08 (0.09)	0.06 (0.08)	0.08 (0.09)
Continuity of care	0.10 (0.11)	0.12 (0.12)	0.11 (0.11)	0.08 (0.09)	0.10 (0.10)
Total health and social care costs	0.03 (0.08)	0.06 (0.09)	0.06 (0.09)	0.08 (0.08)	0.07 (0.09)

Note: Numbers in parentheses (n) by stakeholders indicate the number of participants included in the weight elicitation study

- **Appendix 3:** Graph common support



Common Support of the propensity scores when matching includes the baseline values of the following variables: age, gender, marital status, living situation, educational level, smoking status, and total health and social care costs (minus medication) of three months prior to baseline as a proxy for severity

Appendix 4: Predicted mean performance scores of all outcome measures on their natural scale

Outcomes measures	**T0#**		**T1**		**T2**	
	CCFE (n=222)	**UC (n=162)**	**CCFE (n=172)**	**UC (n=129)**	**CCFE (n=156)**	**UC (n=113)**
Health & Well-being						
Physical functioning (0-15) ^	4.383	4.383	5.126	4.653	5.715	5.328
Psychological well-being (0-100)	71.351	71.351	70.707	71.010	69.998	71.256
Enjoyment of life (1-4)	2.820	2.820	2.917	2.741	2.874	2.839
Social relationships & participation (7-35) ^	9.167	9.167	9.273	9.579	9.424	9.779
Resilience (6-30)	19.317	19.317	19.299	19.108	19.347	19.426
Experience of care						
Person-centeredness (0-18)	11.729	11.729	12.828	11.353	13.060	11.727
Continuity of care (1-5)	3.672	3.672	3.771	3.653	3.833	3.704
Costs						
Total health and social care costs ^	5453	5453	10858	10199	22676	20659
Additional frail elderly-specific outcomes						
Autonomy (7-35)	22.284	22.284	21.785	22.306	21.790	22.834
Burden of medication (0-10) ^	2.088	2.088	2.221	1.878	2.263	1.480

 # Note that the intervention coefficient was included in the calculation of the mean performance scores in the control group, assuming the control group had the same baseline scores as the intervention group to make it possible to directly compare the intervention and control group. CCFE=Care Chain Frail Elderly. UC=Usual care.

Appendix 5: Comparison of baseline characteristics and outcome scores of dropouts versus non-dropouts

Baseline characteristics	**Across CCFE and UC**			**CCFE separately**			**UC separately**		
	Dropouts (n=115)	**Non-dropouts (n=269)**	**P-value**	**Dropouts (n=66)**	**Non-dropouts (n=156)**	**P-value**	**Dropouts (n=49)**	**Non-dropouts (n=113)**	**P-value**
Intervention group	66 (57%)	156 (58%)	0.913	-	-	-	-	-	-
Age	85.00	83.56	0.036*	83.91	83.21	0.449	86.47	84.05	0.016*
Gender (female)	61%	65%	0.434	62%	64%	0.779	59%	66%	0.381
Living situation			0.926			0.842			0.711
• Independent, alone	57%	57%		53%	54%		63%	60%	
• With other(s)	43%	43%		47%	46%		37%	40%	
Marital status			0.162			0.149			0.734
• Single, never married	1%	5%		0%	5%		2%	4%	
• Married or living together	41%	44%		44%	45%		37%	42%	
• Widow(er)	53%	45%		52%	42%		55%	49%	
• Divorced	5%	7%		5%	8%		6%	4%	
Educational level			0.923			0.839			0.981
• Low	70%	71%		68%	71%		71%	70%	
• Middle	19%	17%		23%	19%		14%	15%	
• High	11%	12%		9%	10%		14%	15%	
Smoking (yes)	15%	11%	0.269	18%	13%	0.299	10%	8%	0.641

Appendix 5: Continued

Baseline characteristics	Across CCFE and UC			CCFE separately			UC separately		
	Dropouts (n=115)	Non-dropouts (n=269)	P-value	Dropouts (n=66)	Non-dropouts (n=156)	P-value	Dropouts (n=49)	Non-dropouts (n=113)	P-value
Baseline outcome scores									
Physical functioning (0-15) ^	5.18	4.10	0.001*	4.89	4.17	0.081	5.56	4.02	0.003*
Psychological well-being (0-100)	69.37	72.39	0.145	68.06	72.74	0.092	71.17	71.89	0.815
Enjoyment of life (1-4)	2.81	2.89	0.351	2.74	2.85	0.349	2.90	2.95	0.725
Social relationships and participation (7-35) ^	8.86	8.55	0.461	9.48	9.03	0.451	8.00	7.88	0.831
Resilience (6-30)	18.78	19.38	0.212	18.97	19.46	0.448	18.52	19.27	0.298
Autonomy (7-35)	21.81	22.35	0.245	21.89	22.45	0.386	21.69	22.21	0.452
Person-centeredness (0-18)	12.29	11.97	0.473	11.86	11.66	0.730	12.88	12.4	0.481
Continuity of care (1-5)	3.70	3.72	0.838	3.65	3.69	0.743	3.78	3.77	0.939
Burden of medication (0-10) ^	2.17	2.24	0.809	2.12	2.07	0.908	2.22	2.47	0.604
Total costs 3 months prior to the study (€) ^	5,989	5,112	0.247	5,797	5,308	0.635	6,248	4,842	0.208

*^ Higher score indicating worse performance. * p<0.05. CCFE=Care Chain Frail Elderly. UC=Usual care.*

Appendix 6a: Value scores in the Multi-Criteria Decision Analysis at 6 months without costs

Outcome measures			**Patients**		**Partners**		**Professionals**		**Payers**		**Policymakers**	
	Standardised performance score#		**Weighted score**		**Weighted score**		**Weighted score**		**Weighted score**		**Weighted score**	
	CCFE	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**	**CCFE**	**UC**
Physical functioning	0.672	0.740	0.111	0.122	0.079	0.087	0.085	0.094	0.103	0.114	0.101	0.111
Psychological well-being	0.706	0.709	0.124	0.124	0.113	0.113	0.134	0.134	0.140	0.140	0.114	0.114
Enjoyment of life	0.729	0.685	0.173	0.162	0.194	0.182	0.169	0.159	0.192	0.181	0.172	0.162
Social relationships and participation	0.718	0.696	0.059	0.057	0.069	0.067	0.083	0.081	0.079	0.076	0.077	0.075
Resilience	0.711	0.704	0.110	0.109	0.106	0.105	0.097	0.096	0.086	0.085	0.107	0.106
Person-centeredness	0.749	0.663	0.062	0.055	0.064	0.056	0.063	0.056	0.049	0.044	0.064	0.057
Continuity of care	0.718	0.696	0.074	0.072	0.092	0.089	0.083	0.081	0.063	0.061	0.077	0.075
Overall value scores			**0.712**	**0.701**	**0.715**	**0.698**	**0.714**	**0.700**	**0.713**	**0.701**	**0.713**	**0.700**

colour scheme ranges from red (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care.

Appendix 6b: Value scores in the Multi-Criteria Decision Analysis at 12 months without costs

Outcome measures			Patients		Partners		Professionals		Payers		Policymakers	
	Standardised performance score#		Weighted score		Weighted score		Weighted score		Weighted score		Weighted score	
	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC
Physical functioning	0.682	0.731	0.112	0.121	0.080	0.086	0.086	0.092	0.105	0.113	0.103	0.110
Psychological well-being	0.701	0.713	0.123	0.125	0.112	0.114	0.133	0.135	0.139	0.141	0.113	0.115
Enjoyment of life	0.711	0.703	0.169	0.167	0.189	0.187	0.165	0.163	0.188	0.185	0.168	0.166
Social relationships and participation	0.720	0.694	0.059	0.057	0.069	0.066	0.083	0.080	0.079	0.076	0.077	0.075
Resilience	0.706	0.709	0.109	0.110	0.105	0.106	0.097	0.097	0.085	0.086	0.106	0.107
Person-centeredness	0.744	0.668	0.061	0.055	0.063	0.057	0.063	0.056	0.049	0.044	0.064	0.057
Continuity of care	0.719	0.695	0.074	0.072	0.092	0.089	0.083	0.080	0.063	0.061	0.077	0.075
Overall value scores			**0.708**	**0.706**	**0.710**	**0.704**	**0.710**	**0.704**	**0.708**	**0.706**	**0.709**	**0.705**

colour scheme ranges from red (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care.

Chapter

6

Moving beyond QALYs in elderly care: how can Multi-Criteria Decision Analysis complement Cost-Effectiveness Analysis in local level decision-making

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ABSTRACT

Objectives: To investigate how multi-criteria decision analysis (MCDA) could complement cost-effectiveness analysis (CEA) to support investment decisions in elderly care at local level.

Methods: We used an integrated elderly care programme in the Netherlands as a case study to demonstrate the application of both methods. In a 12-month quasi-experimental study (n=384), data on the following outcome measures were collected: Quality-Adjusted Life Years (QALYs) (CEA) and physical functioning, psychological well-being, social relationships and participation, enjoyment of life, resilience, person-centeredness, continuity of care, and costs (MCDA). We performed regression analysis on inversed probability weighted data and controlled for potential confounders to obtain a double robust estimate of the outcomes. Probabilistic sensitivity analyses determined uncertainty for both methods.

Results: The integrated elderly care programme was not likely (i.e., 36%) to be cost-effective according to the CEA (ICER: € 88,249 from a societal perspective) using the conventional Dutch willingness-to-pay threshold (i.e., €50,000). The MCDA demonstrated that informal caregivers and professionals slightly preferred the intervention over usual care, driven by enjoyment of life and person-centeredness. Patients did not prefer either the intervention or usual care, whereas payers and policymakers slightly preferred usual care, mainly due to higher costs of the intervention.

Conclusion: MCDA could provide local decision-makers with a broader measurement of effectiveness by including outcomes beyond health and longevity and the preferences of multiple stakeholders. This additional information could foster the acceptability and implementability of cost-effective innovations in elderly care.

INTRODUCTION

In cost-effectiveness analysis (CEA), the value of a new intervention versus an alternative is expressed as the additional costs it takes to get an additional unit of health, where the unit of health generally is a quality-adjusted life year (QALY).[1] When the cost-per-QALY ratio is below a certain threshold value that reflects the opportunity costs of a QALY or the willingness-to-pay for a QALY, the intervention is deemed to have added value.[2] This metric is likely to be insufficient to capture the value created by innovations in elderly care, because in practice, the quality-adjustment focuses on health-related quality of life.[3,4] But the main aim of integrated care for frail elderly may not be to improve health, but to support the elderly in ageing-in-place and improve well-being.[5,6] Many elderly care programmes also aim to improve the process of delivering care to the elderly and remove inefficiencies.[7-9] One can measure these additional elements of value and take them into account as contextual factors in support of a deliberate decision-making process, but an alternative approach is to weigh the attributes that matter in value assessment in a multi-criteria decision analysis (MCDA) to better inform that process.

A CEA is done primarily to inform national or regional level decision-making on services to include in the benefit package, generally considered as health technology assessment (HTA) for formulary inclusion.[10] A cascade of decision contexts follows next, from local decision-making about the services a health care organisation plans to offer (guided by clinical guidelines, care pathways and protocols), down to shared clinical decision-making to select the best therapeutic option for an individual. Information on the costs per QALY gained is likely to be insufficient in these lower-level decision-contexts.[11] At these levels, decision-makers need additional information, particularly on the extent to which the goals of an intervention in their specific context were reached. Hence, there is a need for augmented CEA or MCDA that take values beyond the QALY into consideration, particularly in elderly care as innovations in this area include health and social care services.[12-15]

Against this background, the aim of our study was to investigate how MCDA could complement cost-effectiveness analysis (CEA) to support investment decisions in elderly care at local level. We applied both methods to evaluate an integrated elderly care programme, called the Care Chain Frail Elderly (CCFE), in the Netherlands.[16] This responds to the call of ISPOR's (The Professional Society for Health Economics and Outcomes Research) taskforce on MCDA for more head-to-head comparisons of both approaches.[17]

METHODS

Setting and decision context

The study was part of the EU-funded Sustainable Integrated Care Models for Multi-Morbidity Delivery, Financing And Performance (SELFIE) project. One of the aims of this project was to strengthen the evidence-base of integrated care programmes for individuals with multi-morbidity by using a comprehensive evaluation approach, such as MCDA.[18] The case study that was selected concerns the Dutch integrated care programme, the CCFE.[16] It was chosen to demonstrate the additional value of MCDA, because the aim of the care programme reaches beyond health, and because the programme combines health and social care. The decision context for the CCFE was related to providing evidence on the effectiveness of the programme to the professionals providing the programme and the health insurers reimbursing the programme, to support decisions on its continuation, reimbursement and potential upscaling of the programme within the current region and beyond. Therefore, the CCFE was compared to usual care.

Intervention

The CCFE has been described in detail elsewhere.[16] The programme targets the frailest of community-dwelling elderly by integrating services across health and social care and builds a network of support around the patient, aiming to improve their physical, mental, social health and well-being and experience with care. Ideally, this also reduces secondary care and prevents or postpones residential long-term care admissions and thereby costs. A core team of a general practitioner (GP), a nurse practitioner specialised in elderly care, and a district nurse of a home care organisation use a case-finding approach to identify potentially frail elderly. The nurse practitioner visits the elderly at home for a comprehensive geriatric assessment to confirm their frailty and make an inventory of their needs, preferences, capabilities, and resources. Thereafter, a multidisciplinary team meeting is held inviting the health and social care providers involved, a case manager is appointed to coordinate care, and an individual care plan is drafted. The elderly person and his or her informal caregiver are present at the multidisciplinary team meetings and invited to participate actively, and the care programme is financed by a bundled payment in which all care for the frail elderly person provided by the GP-practice is included. Hence, there are no separate consultation-fees for disease-related primary care services provided by the GP-practice.

Study design and data collection

In this 12-month prospective quasi-experimental study, we collected data among elderly newly enrolled in the CCFE (intervention group) and among frail elderly receiving usual care (control group). Elderly in the control group, living in the same region as elderly in the intervention group, were identified by their GP using the same case-finding approach as was applied in the intervention group. Data collection took place at baseline, after 6 and 12 months, and consisted of an extensive questionnaire covering outcomes that span the triple aim of integrated care: improving health and well-being, improving experience with care, and reducing costs(-increase).[19] Trained interviewers visited the frail elderly at home to administer the questionnaire, to ensure that very frail elderly could participate and that questions were well understood by respondents. The medical ethics committee of the Erasmus Medical Centre, the Netherlands reviewed the study protocol and concluded that the rules laid down in the Dutch Medical Research Involving Human Subjects Act do not apply to this research (*MEC-2017-121*). All study participants have provided informed written consent. Data collection took place between April 2017-August 2019.

Outcomes

The main outcome measure in the CEA was QALYs, whereas the main outcome measure in the MCDA was an overall value score, calculated as a weighted sum of eight outcomes measures. The latter included two health outcomes, i.e., physical functioning and psychological well-being, three well-being outcomes, i.e., social relationships and participation, enjoyment of life, and resilience, two care-process outcomes, i.e., person-centeredness, continuity of care, and total costs. These outcomes were selected based on literature reviews, workshops with representatives from five stakeholder groups, and focus groups with individuals with multi-morbidity, and measured with validated questionnaires (**Appendix Table 1**).[20]

Costs

In both the CEA and the MCDA, costs were calculated from a societal perspective as well as a healthcare perspective. The healthcare perspective included the following cost categories: care programme costs, homecare, long-term care admissions, hospital admissions, emergency room visits, outpatient day-care activities, medical specialist care, paramedical care, general practitioner, and medication. The societal perspective additionally included informal care costs. Resource utilisation data were obtained with the iMTA-Medical Consumption Questionnaire, using a 3-month recall period.[21] Unit costs were largely based on reference prices from the Dutch Costing Manual which were converted into 2018 prices using Consumer Price Index provided by Statistics Netherlands.[22,23] Medication prescriptions were obtained from GP-information systems. In the Netherlands, GPs have formed Care Groups, that negotiate with the health insurers about the bundled payment for integrated chronic care programmes.[16] CCFE programme costs were retrieved from these Care Groups.[16]

Weighting outcomes

In the CEA, the weights used to perform the health-related quality of life (HRQoL) adjustments in the QALY calculation were based on the Dutch EQ-5D-5L tariff.[24] These weights were derived from the general population and only pertain to health outcomes. In the MCDA, weights were derived from five different stakeholder groups, i.e., persons with multi-morbidity, informal caregivers, professional care providers, payers, and policymakers, resulting in five different weight sets, see **Appendix Table 2**. These represent the most important stakeholders whose views are relevant to inform decision-making on the CCFE. To obtain these weights a discrete choice experiment (DCE) in 724 Dutch respondents was conducted, the results of which were published elsewhere.[25]

Statistical analysis

Data analysis was performed according to the intention-to-treat principle. As this was a non-randomised study, we used inverse probability weighting (IPW) to increase comparability between intervention and control group.[25-28] To calculate the inverse probability weight, the following variables were included in the propensity score: age, gender, marital status, living situation, educational level, smoking status, and costs three months prior to baseline as a proxy for frailty. Out of several matching techniques, IPW was chosen because it led to the best matching statistics, i.e., Rubin's B was below 25 and Rubins' R between 0.5-2. A detailed description of this method was given in a previous paper.26

A double robust estimate of the outcomes was obtained by performing regression analysis on inversed probability weighted data and controlling for potential confounders.[29] Weighted linear mixed models were used to predict the mean scores of all outcome measures in both groups at each time point, assuming the control group had the same baseline score as the intervention group. We included a random intercept at individual level and corrected for time, intervention, the interaction between time and intervention, age, gender, marital status, living situation, educational level, and smoking status.

All analyses were performed in Stata 16.1.

CEA

We calculated the number of QALYs for each patient as the area under the predicted utility curve. The incremental cost-effectiveness ratios (ICERs) were calculated as the mean difference in total costs divided by the mean difference in QALYs.

MCDA

All predicted outcome scores used in the MCDA were standardised on a 0-1 scale using relative standardisation:

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

Where x is the predicted mean score on the natural scale, *a* is the intervention group, *b* is the control group, and *j* is outcome measure. For all outcomes in the MCDA, a higher score indicates better performance. Hence, x is replaced by $1/x$ for outcomes where a higher score on the natural scale indicates a worse performance (e.g., costs). Subsequently, all standardised outcome scores were weighted according to each of the five stakeholder perspectives and summed to gain overall value scores for both the intervention and control group.

Uncertainty analysis

In the CEA, uncertainty was illustrated by the CE-plane and the Cost-Effectiveness Acceptability Curve (CEAC) using the ICERs of 10,000 bootstrap replications. The CEAC represents the proportion of replications in which the ICER is lower than the threshold value of the willingness-to-pay for a QALY. The Dutch threshold is determined by the burden of disease expressed in QALYs lost due to disease as a proportion of quality-adjusted life expectancy of the age- and gender-matched general population (i.e., proportional shortfalls).[30] The proportional shortfall was estimated to be 0.66, which indicates that a threshold value of € 50,000 per QALY should be used.[31]

In the MCDA, uncertainty was illustrated with the Conditional Multi-attribute Acceptability Curve (CMAC). We used Monte-Carlo simulations to assess the joint uncertainty of the preference weights and outcome scores.[32,33] We used the Cholesky decomposition to obtain 10,000 replications of the weights and outcome scores. For each replication, overall value scores were calculated, as previously described, to determine how often the CCFE was preferred over usual care, and to present the 95% confidence interval around the overall value scores. To draw the CMAC, we calculated the probability the CCFE is the preferred alternative (i.e., has a higher overall value score) while the budget remains below a set threshold. This threshold represents the available budget, given a certain target population size. We used a target population of 2,000 frail older persons, which reflects approximately 100 GP’s that would implement the CCFE (average general practice size: 2095 patients[34] and the CCFE targets approximately 1% of them).[16]

Deliberation

An integral part of performing an MCDA is the deliberation on the results.[18] Representatives from the five stakeholder groups discussed the outcome measures included in the study and the weights resulting from the DCE. MCDA results were reflected upon by professionals and payers directly involved in the care programme, for example GPs and representatives from the care groups and health insurance company.

RESULTS

Characteristics of the sample

In total, 384 frail elderly participated in the study, 222 in the CCFE group and 162 receiving usual care. Their mean age was 83.4 years and 83.6 years, respectively (**Table 1**). Women were overrepresented in both groups. There are very little differences in baseline characteristics between groups, except for social relationships and participation and person-centeredness, where the intervention group scores a bit worse. In both the intervention and control group the loss to follow-up was 30% after 12-months. Main reasons of loss-to-follow-up included death, burden of study participation, and cognitive incapacity.

Cost-effectiveness analysis

Over a 1-year period, the number of QALYs was 0.023 higher in the CCFE group than in the usual care group (**Table 2**). When adopting a healthcare perspective, the total costs in the CCFE group were on average € 14,418 for each frail elderly person over a 1-year period while these costs were € 12,785 in the control group. As a result, the incremental costs of the CCFE were € 1,633 from a healthcare perspective, and € 2,017 from a societal perspective. A specification with all cost categories can be found in **Appendix Figure 1**. The ICERs for the CCFE were € 71,460 and € 88,249, respectively.

Table 1. Baseline characteristics and baseline values of outcome measures

Baseline characteristics	**CCFE (n = 222)**	**UC (n = 162)**	**P-value**
Age, mean (SD)	83.4 (6.3)	83.6 (5.9)	0.768
Female (%)	63.5	64.8	0.780
Marital status (%)	3.6	2.6	0.973
• Single, never married	44.6	45.0	
• Married or living together	44.6	47.1	
• Widow(er)	7.2	5.4	
• Divorced			
Living situation (%)	54.1	55.2	0.818
• Independent, alone	46.0	44.8	
• With others			
Educational level (%)	70.3	73.3	0.986
• Low	20.3	14.3	
• Medium	9.5	12.4	
• High			
Current smoker (%)	14.4	13.8	0.863
Baseline values of outcome measures, mean (SD)			
Physical functioning (0-15) ^	4.4 (2.8)	4.5 (3.2)	0.597
Psychological well-being (0-100)	71.4 (18.9)	70.6 (18.2)	0.673
Health-related quality of life (0-1)	0.6 (0.3)	0.6 (0.3)	0.785
Enjoyment of life (1-4)	2.8 (0.8)	2.9 (0.9)	0.138
Social relationships and participation (7-35) ^	9.2 (4.1)	8.2 (3.2)	0.004*
Resilience (6-30)	19.3 (4.4)	19.0 (4.3)	0.392
Autonomy (7-35)	22.3 (4.3)	21.9 (4.1)	0.309
Burden of medication (0-10) ^	2.1 (2.8)	2.5 (2.8)	0.086
Person-centeredness (0-18)	11.7 (3.9)	12.7 (3.9)	0.010*
Continuity of care (1-5)	3.7 (0.8)	3.8 (0.7)	0.067
Total costs 3 months prior to the study (€) ^	5,453 (6,994)	5,631 (6,946)	0.789

*CCFE=Care Chain Frail Elderly; UC=Usual Care; SD=Standard Deviation. *=p<0.05. ^Higher score indicating worse performance. Note: This table presents the baseline characteristics and outcome scores after IPW. The flowchart of patient inclusion and the results of IPW have been presented elsewhere*[25]

• **Table 2.** QALYs, costs and ICERs from a healthcare- and societal perspective

	CCFE (n=156)	Usual care (n=113)	Incremental (95% CI^)
Total QALYs	0.665	0.642	0.023 (-0.071 – 0.122)
Total costs			
Healthcare perspective	€ 14,418	€ 12,785	€ 1,633 (-1,776 – 5,042)
Societal perspective	€ 22,676	€ 20,659	€ 2,017 (-2,361 – 6,395)
ICER - Healthcare perspective			€ 71,460
ICER - Societal perspective			€ 88,249

Abbreviations: CCFE=Care Chain Frail Elderly. CI=Confidence Interval. QALYs=Quality-Adjusted Life-Years. ICER=Incremental Cost-Effectiveness Ratio. ^95% CI based on PSA.

The cost-effectiveness plane shows that the majority of the ICERs (67% from a healthcare perspective and 68% from a societal perspective) appeared in the northeast quadrant, indicating that the CCFE was more effective than usual care but also more costly (**Figure 1a and 1b**). From a healthcare perspective, 18% of the bootstrap replications fell within the southeast quadrant of the plane, indicating that the CCFE was the dominant strategy, i.e., less costly and more effective (**Appendix Figure 2**). The probability that the CCFE was cost-effective at a willingness-to-pay of € 50,000 per QALY at 12 months was 42% from a healthcare perspective. From a societal perspective, the probability was slightly lower, that is, 36%.

Multi-Criteria Decision Analysis

The predicted scores and standardised scores on all outcome measures at 12 months are presented in **Table 3**. Largest differences in performance scores between the CCFE and usual care were found in the scores for person-centeredness in favour of the CCFE (CCFE: 0.744 vs UC: 0.668) and total costs from a healthcare perspective in favour of usual care (0.663 vs 0.748).

● **Figure 1a and 1b.** Cost-effectiveness planes and cost-effectiveness acceptability curves obtained by bootstrap replications, from a societal perspective

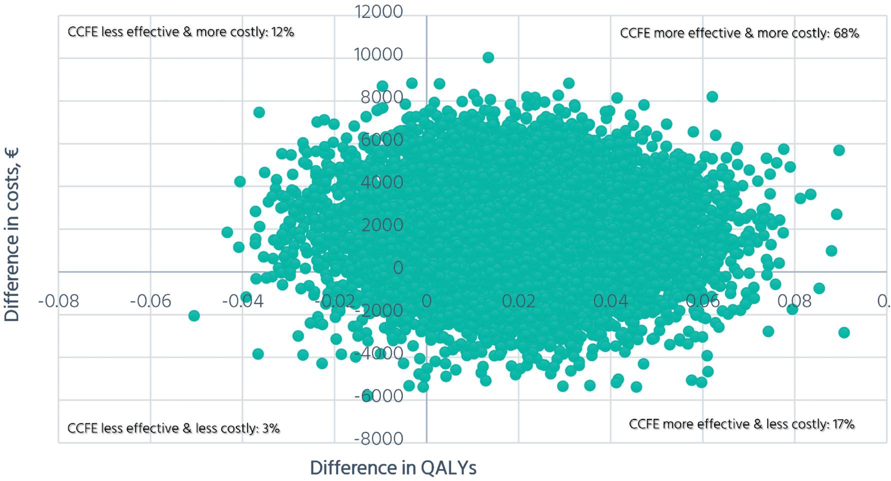


Figure 1a. Cost-effectiveness plane from a societal perspective

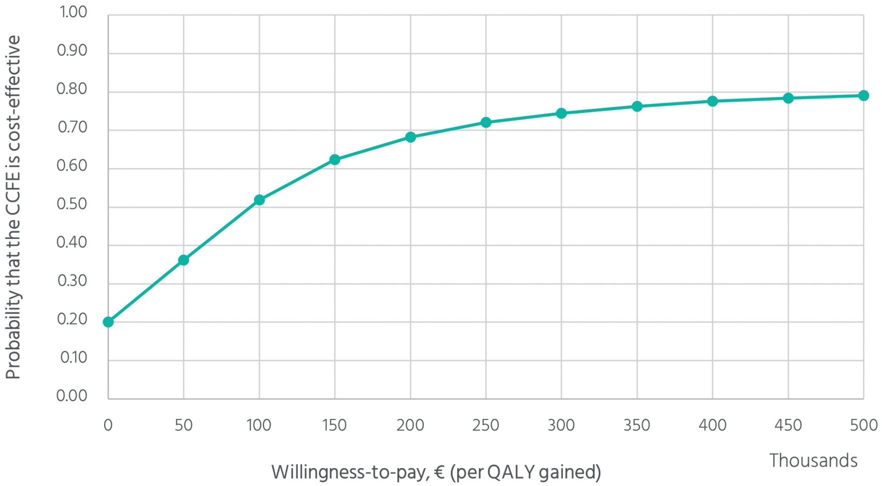


Figure 1b. Cost-effectiveness acceptability curve from a societal perspective

Table 3. Predicted and standardised scores at 12 months for the CCFE and usual care

Outcomes measures	Predicted scores		Standardised scores	
	CCFE (n=156)	Usual care (n=113)	CCFE	Usual care
Health & Well-being				
Physical functioning (0-15)	5.715^	5.328^	0.682	0.731
Psychological well-being (0-100)	69.998	71.256	0.701	0.713
Enjoyment of life (1-4)	2.874	2.839	0.711	0.703
Social relationships & participation (7-35)	9.424^	9.779^	0.720	0.694
Resilience (6-30)	19.347	19.426	0.706	0.709
Experience of care				
Person-centeredness (0-18)	13.060	11.727	0.744	0.668
Continuity of care (1-5)	3.833	3.704	0.719	0.695
Costs				
Total health and social care costs (€) – healthcare perspective	14,418^	12,785^	0.663	0.748
Total health and social care costs (€) – societal perspective	22,676^	20,659^	0.673	0.739

^ Higher score indicating worse performance. CCFE=Care Chain Frail Elderly.

Table 4 presents the MCDA table with the weights, weighted scores and summed overall value scores for the CCFE and usual care. It includes the costs from a societal perspective. In all stakeholder groups, enjoyment of life received the highest relative importance-weight (range 0.22-0.25), followed by psychological well-being (range 0.15-0.18). Total costs received the lowest relative importance-weight (range 0.03-0.07) according to patients, partners, professionals, and policymakers. Payers attached the least importance to person-centeredness (0.06). For patients, the overall value scores did not differ between the CCFE and usual care. The overall value scores for the CCFE were numerically slightly higher than usual care when weighted according to the preferences of partners (0.708 vs 0.706) and professionals (0.714 vs 0.713) and slightly lower when weighted according to the preferences of payers (0.697 vs 0.702) and policymakers (0.706 vs 0.708). The latter was mainly driven by a lower performance on physical functioning and higher costs for the CCFE. Confidence intervals around the value scores of the two groups largely overlapped. The MCDA table with costs from a healthcare perspective can be found in **Appendix Table 3**, showing that the overall value scores for the CCFE and usual care were equal according to patients, informal caregivers and professionals. Payers and policymakers still preferred usual care.

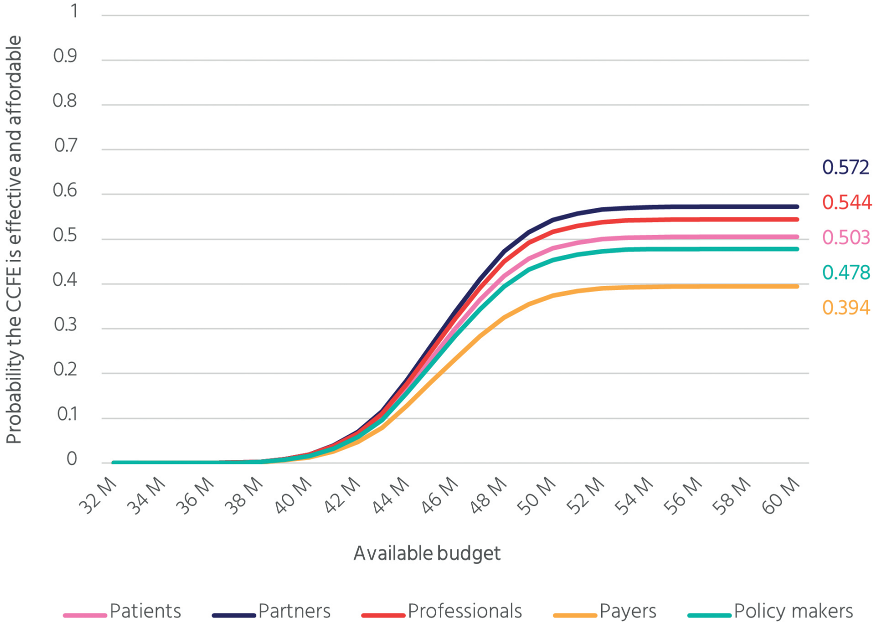
- **Table 4.** MCDA table: Weights, weighted performance scores, and overall value scores for the CCFE and usual care from a societal perspective

Outcome measures	**Patients**			**Partners**			**Professionals**			**Payers**			**Policy makers**		
		Weighted score			**Weighted score**			**Weighted score**			**Weighted score**			**Weighted score**	
	Weight	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**
Physical functioning	0.16	0.109	0.117	0.11	0.075	0.080	0.12	0.082	0.088	0.14	0.095	0.102	0.14	0.095	0.102
Psychological well-being	0.17	0.119	0.121	0.15	0.105	0.107	0.18	0.126	0.128	0.18	0.126	0.128	0.15	0.105	0.107
Enjoyment of life	0.23	0.164	0.162	0.25	0.178	0.176	0.22	0.157	0.155	0.24	0.171	0.169	0.22	0.157	0.155
Social relationships and participation	0.08	0.058	0.056	0.09	0.065	0.062	0.11	0.079	0.076	0.10	0.072	0.069	0.10	0.072	0.069
Resilience	0.15	0.106	0.106	0.14	0.099	0.099	0.13	0.092	0.092	0.11	0.078	0.078	0.14	0.099	0.099
Person-centeredness	0.08	0.060	0.053	0.08	0.060	0.053	0.08	0.060	0.053	0.06	0.045	0.040	0.08	0.060	0.053
Continuity of care	0.10	0.072	0.069	0.12	0.086	0.083	0.11	0.079	0.076	0.08	0.058	0.056	0.10	0.072	0.069
Total costs - societal perspective	0.03	0.020	0.022	0.06	0.040	0.044	0.06	0.040	0.044	0.08	0.054	0.059	0.07	0.047	0.052
Overall value scores		0.707	0.707		0.708	0.706		0.714	0.713		0.698	0.702		0.706	0.707
95% CI^		0.696 - 0.717	0.697 - 0.717		0.697 - 0.718	0.695 - 0.716		0.697 - 0.717	0.695 - 0.716		0.695 - 0.717	0.697 - 0.719		0.695 - 0.717	0.696 - 0.718
% CCFE>UC		50%			57%			54%			39%			48%	

^ *based on Monte Carlo simulation; Colour scheme ranges from white (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care. CI=Confidence Interval.*

The CMAC (**Figure 2**) shows the probability of the CCFE being effective and affordable from a societal perspective given a certain target population and available budget. The CMAC that considers the healthcare perspective can be found in **Appendix Figure 3**. The probability that the CCFE is effective and affordable (compared to usual care) increases to a maximum of 50% for patients, 54% for professionals, and 57% for partners at a budget of € 56,000,000 for a 1-year period for 2,000 frail elderly persons, or € 28,000 per person. For payers and policymakers, the probability of being effective and affordable is higher for usual care.

● **Figure 2.** Conditional Multi-Attribute Acceptability Curves from a societal perspective presenting the probability of the CCFE to be accepted as the preferred alternative over different levels of budget available to be allocated to either the CCFE or usual care given a target population of 2,000 frail elderly persons



DISCUSSION

In the current study we investigated how MCDA could complement CEA to aid local-level decision-making in elderly care by applying both methods to a person-centred integrated elderly care programme, the CCFE. According to CEA and the associated CEAC, the programme was not likely to be cost-effective at a willingness-to-pay threshold of € 50,000. The MCDA and associated CMAC demonstrated the CCFE is potentially equally effective and affordable as usual care. Therefore, the investment decision based on the two approaches could differ. Using CEA, the CCFE would be very unlikely to be investable, but using MCDA decision-makers may choose to favour the intervention, because given the uncertainty they could improve enjoyment of life (the most important criterion for all stakeholder groups) and promote person-centred care, which is in line with national policy guidance.

MCDA results could support decision-makers, especially on a local level. Whereas on a national level it is decided to put focus on integrated elderly care, the further interpretation of how this care should look like is delegated to the regional level. In the Netherlands, these interventions are often developed by care providers, but the decision to reimburse new types of care lays with the health insurers, whereas in the UK the decision-makers could be the Integrated Care Systems[35] and in Australia the Primary Health Networks.[36] However, decision-makers could benefit from additional information as improvements in HRQoL are difficult to measure and/or achieve in elderly care.[5] Furthermore, our findings showed that HRQoL (as measured by physical functioning and psychological well-being) did not receive the highest relative importance by any stakeholder group, which questions the suitability of QALYs, and therefore CEA in this context. MCDA could provide such additional information. First, in MCDA a broad set of outcome measures can be taken into account, for example covering the triple aim. Second, in MCDA it is possible to incorporate multiple stakeholder perspectives by means of different weight sets. In CEA it is not easily possible to include other perspectives on the value of health outcomes than that of the general population – as value sets are derived from the general population. Third, MCDA is useful in comparing and/or ranking new initiatives in elderly care, as programmes may focus on various aims, e.g., improving social participation versus improving experience with care. Lastly, MCDA can facilitate the choice between many different elderly care initiatives, as are often presented to local level decision-makers.

Comparison with other research

There are two other studies that investigated how MCDA relates to CEA and whether it can alter the decision of continuation, reimbursement, and upscaling of innovations, albeit focusing on other types of interventions and target groups. The first study used the weights elicited in the SELFIE study to perform an MCDA of 'Primary Care Plus', a new model care in which GPs could refer patients to medical specialists located at a primary care site.[37] However, they did not collect performance on the full range of outcome measures, and weights needed to be rescaled. This may have led to the comparable results of CEA and MCDA, and the decision to recommend Primary Care Plus as it was dominant over usual care in both methods. The second study, an evaluation of interventions for knee osteoarthritis[38], investigated to which explicit extent stakeholders' preferences elicited in the MCDA were consistent with the more implicit trade-offs made in CEA. Hence, they used the same outcomes in MCDA and CEA, thereby ignoring the potential of MCDA to adopt a broader view of a program's effectiveness. They found a good correlation in intervention ranking between MCDA and CEA, which was to be expected as both approaches included the same outcomes. Even though in both studies the decision was not altered, the richness of information MCDA provided could shape up decisions, which could be altered as innovations are evolving and need close monitoring and "fine-tuning", rather than a one-off decision.

The latter study did find differences in the perception of costs between the models, as low-cost interventions were consistently ranked higher in the CEA than in the MCDA, and vice versa. Hence, one of their conclusions is to exclude 'cost' as an outcome measure from the MCDA. This is in accordance with recent literature that has discussed the potential of MCDA in health technology assessment, but emphasises the need for advancement in robust methodology.[17,39 -44] One of those methodological challenges is whether to include cost as a criterion. An advantage of including costs is that the relative importance stakeholders attach to costs, is determined by deliberately making trade-offs between costs and the other outcomes. Others argue that the opportunity costs are then not addressed adequately, as it is unrealistic to assume that respondents can fulfil this task.[45] Therefore, outcome measures like costs, resource use, and cost-effectiveness, should not be included in the MCDA. Accordingly, we performed the MCDA without costs by rescaling the weight sets, see **Appendix Table 4.** Results showed that the CCFE is then preferred by all stakeholder groups. However, now opportunity costs for the new composite benefit score created with the MCDA need to be estimated. This is similar to CEA, where the opportunity costs for 1 additional QALY are estimated. This also requires determining cost-effectiveness thresholds for each composite benefit score, which may not be feasible, especially in decision-making on a local level, as outcome measures in the MCDA are context-specific and not generalisable. Comparisons between several diseases would be difficult, and it would be up to the policymaker to decide whether there is enough budget. Additionally, we showed that the CMAC could then aid in allocating the budget that is available for a certain target population. Where in CEA budget impact is dealt with as a separate criterion, apart from the CEAC, in MCDA affordability is already included in the CMAC.

Implications

The choice of which method to consult to aid your decision, depends on the decision context. CEA is recommended to inform decisions pertaining to the benefit package on a national level, based on the costs/QALY and existing willingness-to-pay thresholds. Yet, in local-level decision-making CEA may be insufficient to fully determine the value of interventions. Given the benefits of MCDA, we advocate for an ongoing monitoring framework based on MCDA to be used in investment decisions in elderly care where a QALY is not informative to decision-makers. As weight sets were already derived from five stakeholder groups, only data regarding the performance scores need to be collected to carry out the MCDA. Using routinely collected data, the intervention could be monitored and continuously adapted based on results. This MCDA-framework could then improve the acceptability of innovations by a) the patients as they can get information on outcomes they can comprehend, such as enjoyment of life and person-centeredness, b) health- and social care professionals, as they see the reflection of their work on intermediate outcomes such as person-centeredness which helps to stay motivated, c) managers/commissioners/payers, as they can monitor the performance of ongoing innovations on short and medium term outcomes while keeping services within budgets.

Strengths & limitations

One of the main strengths of this study is the comprehensive data collection among a difficult to study target group. Although this could be challenging in implementing an MCDA-framework, we demonstrated in our case study that it is feasible by closely collaborating with the care providers and embedding the evaluation study early in the implementation of the integrated care programme. Trained interviewers administered the questionnaire during home visits, enabling frail elderly that would otherwise not be capable of filling in such an extensive questionnaire, to participate. However, this additional data collection may be less of a burden in the future as PROMS and PREMs are increasingly becoming part of routinely collected data.[46] Another difficulty of implementing an MCDA-framework lies in the comparative opportunities between interventions. To achieve this, there needs to be consensus about which outcomes to include in the MCDA-framework. In our study this consensus was reached by involving all relevant stakeholder groups during the selection of outcome measures.

Our study has not been able to demonstrate the full potential of MCDA. Firstly, the effect of the intervention was small explaining why differences between MCDA and CEA were not very distinct. If the programme had larger effects reflected in performance scores on multiple outcomes, differences between both methods may have been more explicit. Secondly, we only compared the CCFE with usual care instead of assessing multiple interventions, thereby not exploiting all possibilities available with MCDA. Thirdly, the differences in weights between the stakeholder groups were not large, which led to small differences in overall value scores.

CONCLUSION

In comparison with CEA, MCDA could provide local decision-makers with a broader measurement of effectiveness by including outcomes beyond health and longevity and the preferences of multiple stakeholders, in a systematic manner. This additional information aids decision-making in elderly and social care – where interventions are generally pointed at improving these broader outcomes – because decisions regarding reimbursement or scaling-up become more transparent and nuanced. This could foster the acceptability and implementability of integrated elderly care innovations.

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SUPPLEMENTARY MATERIAL

- **Appendix Table 1.** Outcome measures and instruments

CEA	**Instrument to measure outcome**	**Scale**
QALYs	EuroQOL 5D-5L[1]	0-1 (best)
MCDA	**Instrument to measure outcome**	**Scale**
Health & well-being		
Physical functioning	Activities of Daily Living (ADL) (Katz-15)[2]	0-15 (worst)
Psychological well-being	Mental Health Inventory (MHI-5)[3]	0-100 (best)
Enjoyment of life	Investigating Choice Experiments for the Preferences of Older People (ICECAP-O)[4]	1-4 (best)
Social relationships & participation	Impact on Participation & Autonomy (IPA), social life and relationships domain[5]	0-28 (worst)
Resilience	Brief Resilience Scale (BRS)[6]	6-30 (best)
Experience of care		
Person-centeredness	Person-centered Coordinated Care Experience Questionnaire (P3CEQ), experience of person-centered care domain[7]	0-18 (best)
Continuity of care	Nijmegen Continuity Questionnaire (NCQ), team and cross boundary continuity domain[8] + Client Perceptions of Coordination Questionnaire (CPCQ)[9]	1-5 (best)
Total costs		
Health, social, and informal care costs	iMTA Medical Consumption Questionnaire[10]	
Medication costs	Prescriptions in patient records extracted from GP information systems	
Bundled payments and chronic care programmes	Care chain information system 'Care2U'	

References Appendix Table 1

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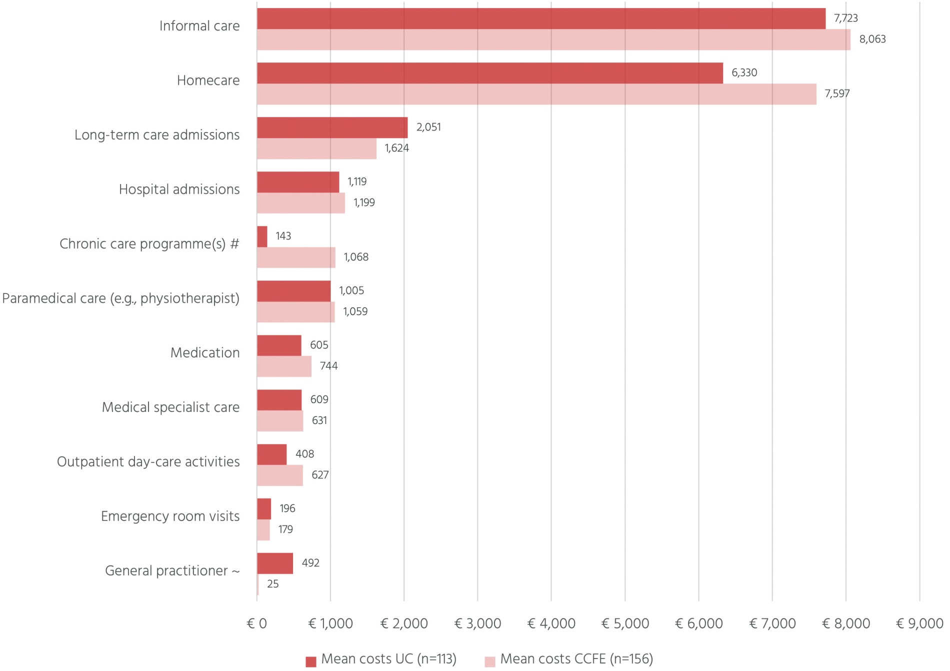
● **Appendix Table 2.** Relative DCE weights (0-1) (SE) of the outcomes used in the MCDA by type of stakeholder

	Patients (n=156)	**Partners (n=158)**	**Professionals (n=155)**	**Payers (n=104)**	**Policymakers (n=151)**
Physical functioning	0.16 (0.15)	0.11 (0.12)	0.12 (0.12)	0.14 (0.13)	0.14 (0.14)
Psychological well-being	0.17 (0.15)	0.15 (0.14)	0.18 (0.16)	0.18 (0.17)	0.15 (0.15)
Enjoyment of life	0.23 (0.18)	0.25 (0.19)	0.22 (0.19)	0.24 (0.23)	0.22 (0.19)
Social participation & relationships	0.08 (0.10)	0.09 (0.11)	0.11 (0.11)	0.10 (0.11)	0.10 (0.10)
Resilience	0.15 (0.13)	0.14 (0.13)	0.13 (0.12)	0.11 (0.10)	0.14 (0.13)
Person-centeredness	0.08 (0.10)	0.08 (0.10)	0.08 (0.09)	0.06 (0.08)	0.08 (0.09)
Continuity of care	0.10 (0.11)	0.12 (0.12)	0.11 (0.11)	0.08 (0.09)	0.10 (0.10)
Total health and social care costs	0.03 (0.08)	0.06 (0.09)	0.06 (0.09)	0.08 (0.08)	0.07 (0.09)

Note: Numbers in parentheses (n) by stakeholders indicate the number of participants included in the weight elicitation study. The relative DCE weight of each of the outcomes was calculated as the contribution of the outcome's best level coefficient (level 3) to the sum of all outcomes' best level coefficients.

- **Appendix Figure 1.** Mean costs after 12 months follow-up



 # Costs for chronic care program(s) is an average estimation which is the same for each respondent in their respective group; ~ Costs for GP-care in the intervention group are largely included in the costs of the chronic care programme. CCFE=Care Chain Frail Elderly. UC=Usual care.

Data table belonging to Appendix Figure 1. Mean costs after 12 months follow-up

	Chronic care program(s) #		Homecare		Long-term care admissions		Hospital admissions		Emergency room visits		Outpatient day-care activities	
	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC
Mean	1068	143	7597	6330	1624	2051	1199	1119	179	196	627	408
Standard deviation	0	0	8474	722	7959	943	3603	281	372	39	2642	174
Median	1068	143	4789	3468	0	0	0	0	0	0	0	0
25%	1068	143	1767	1734	0	0	0	0	0	0	0	0
75%	1068	143	10570	8281	0	0	0	0	268	536	0	0
Interquartile range	0	0	8803	6547	0	0	0	0	268	536	0	0

Cont'd	Medical specialist care		Paramedical care (e.g., phys-iotherapist)		General practitioner ~		Medication		Informal care	
	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC	CCFE	UC
Mean	631	609	1059	1005	25	492	744	605	8063	7723
Standard deviation	789	58	1113	107	34	37	1232	87	12436	1283
Median	459	388	678	469	15	410	481	437	3388	2085
25%	188	188	288	210	15	227	188	210	1129	232
75%	941	829	1549	1707	15	622	911	620	9788	7153
Interquartile range	753	641	1261	1497	0	395	722	411	8659	6921

 # Costs for chronic care program(s) is an average estimation which is the same for each respondent in their respective group; ~ Costs for GP-care in the intervention group are largely included in the costs of the chronic care programme. CCFE=Care Chain Frail Elderly. UC=Usual care.

● **Appendix Figure 2.** Cost-effectiveness planes and cost-effectiveness acceptability curves obtained by bootstrap replications, from a healthcare perspective

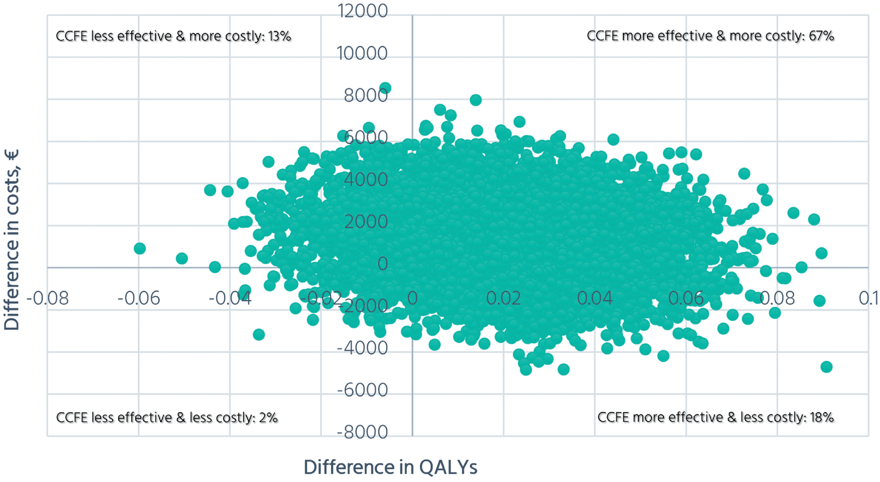


Figure 2a. Cost-effectiveness plane from a societal perspective

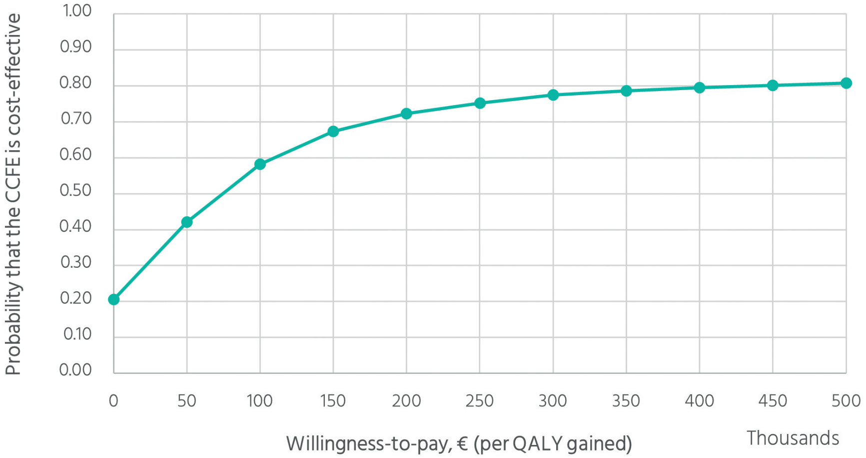


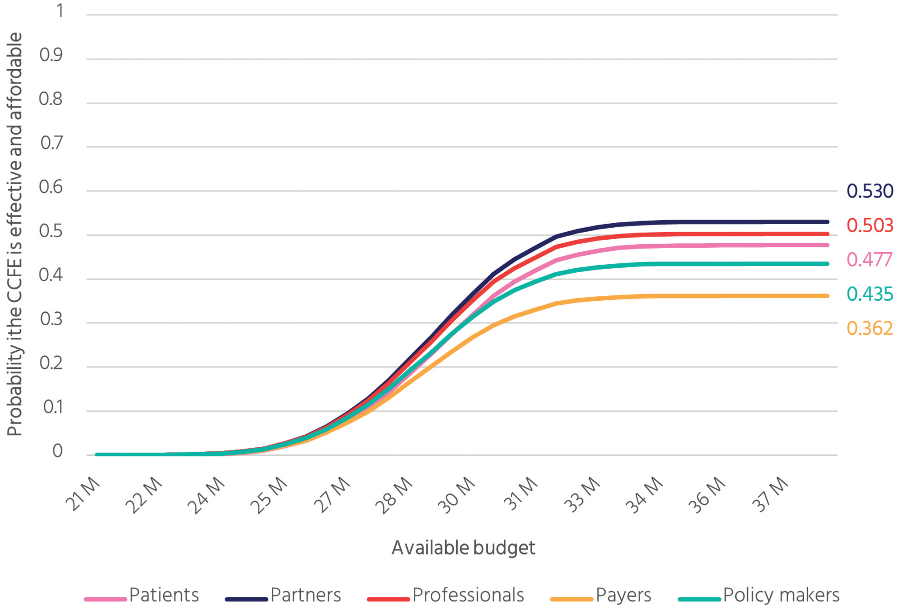
Figure 2b. Cost-effectiveness acceptability curve from a societal perspective

Appendix Table 3. Results of the Multi-Criteria Decision Analysis at 12 months from a healthcare perspective, using DCE weights

Outcome measures	**Patients**			**Partners**			**Professionals**			**Payers**			**Policy makers**		
	Weighted score			**Weighted score**			**Weighted score**			**Weighted score**			**Weighted score**		
	Weight	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**
Physical functioning	0.16	0.109	0.117	0.11	0.075	0.080	0.12	0.082	0.088	0.14	0.095	0.102	0.14	0.095	0.102
Psychological well-being	0.17	0.119	0.121	0.15	0.105	0.107	0.18	0.126	0.128	0.18	0.126	0.128	0.15	0.105	0.107
Enjoyment of life	0.23	0.164	0.162	0.25	0.178	0.176	0.22	0.157	0.155	0.24	0.171	0.169	0.22	0.157	0.155
Social relationships and participation	0.08	0.058	0.056	0.09	0.065	0.062	0.11	0.079	0.076	0.10	0.072	0.069	0.10	0.072	0.069
Resilience	0.15	0.106	0.106	0.14	0.099	0.099	0.13	0.092	0.092	0.11	0.078	0.078	0.14	0.099	0.099
Person-centeredness	0.08	0.060	0.053	0.08	0.060	0.053	0.08	0.060	0.053	0.06	0.045	0.040	0.08	0.060	0.053
Continuity of care	0.10	0.072	0.069	0.12	0.086	0.083	0.11	0.079	0.076	0.08	0.058	0.056	0.10	0.072	0.069
Total costs - healthcare perspective	0.03	0.020	0.022	0.06	0.040	0.045	0.06	0.040	0.045	0.08	0.053	0.060	0.07	0.046	0.052
Overall value scores		0.707	0.707		0.707	0.707		0.714	0.714		0.697	0.702		0.706	0.708
95% CI^		0.696 - 0.717	0.697 - 0.717		0.696 - 0.718	0.695 - 0.717		0.695 - 0.718	0.696 - 0.718		0.692 - 0.716	0.697 - 0.720		0.694 - 0.717	0.696 - 0.719
% CCFE>UC		48%			53%			50%			35%			44%	

^ based on Monte Carlo simulation; # colour scheme ranges from red (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care.

● **Appendix Figure 3.** Conditional Multi-Attribute Acceptability Curves from a healthcare perspective



Appendix Table 4. Results of the Multi-Criteria Decision Analysis at 12 months without costs

Outcome measures	**Patients**			**Partners**			**Professionals**			**Payers**			**Policy makers**		
	Weighted score			**Weighted score**			**Weighted score**			**Weighted score**			**Weighted score**		
	Weight	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**
Physical functioning	0.16	0.112	0.121	0.12	0.080	0.086	0.13	0.086	0.092	0.15	0.105	0.113	0.15	0.103	0.110
Psychological well-being	0.18	0.123	0.125	0.16	0.112	0.114	0.19	0.133	0.135	0.20	0.139	0.141	0.16	0.113	0.115
Enjoyment of life	0.24	0.169	0.167	0.27	0.189	0.187	0.23	0.165	0.163	0.26	0.188	0.185	0.24	0.168	0.166
Social relationships and participation	0.08	0.059	0.057	0.10	0.069	0.066	0.12	0.083	0.080	0.11	0.079	0.076	0.11	0.077	0.075
Resilience	0.15	0.109	0.110	0.15	0.105	0.106	0.14	0.097	0.097	0.12	0.085	0.086	0.15	0.106	0.107
Person-centeredness	0.08	0.061	0.055	0.09	0.063	0.057	0.08	0.063	0.056	0.07	0.049	0.044	0.09	0.064	0.057
Continuity of care	0.10	0.074	0.072	0.13	0.092	0.089	0.12	0.083	0.080	0.09	0.063	0.061	0.11	0.077	0.075
Overall value scores		0.708	0.706		0.710	0.704		0.710	0.704		0.708	0.706		0.709	0.705
95% CI^		0.698 - 0.718	0.695 - 0.716		0.700 - 0.720	0.693 - 0.714		0.700 - 0.720	0.694 - 0.714		0.697 - 0.718	0.695 - 0.716		0.699 - 0.719	0.694 - 0.715
% CCFE>UC		59%			72%			71%			58%			66%	

^ based on Monte Carlo simulation; $^{\#}$ colour scheme ranges from red (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care.

Chapter 7

General discussion: Value-based integrated care in context

GENERAL DISCUSSION

Person-centred integrated care is widely considered a solution to the challenge of maintaining the quality, efficiency, and affordability of healthcare. Evidence from sound evaluations of integrated care interventions is vital to support the efficient allocation of scarce financial, human, and technological resources to the different (often competing) healthcare services. Yet, various studies have shown that the evidence on the (cost-) effectiveness of integrated care programmes is often of poor quality and inconsistent.[1-8] The question arises whether this can be attributed to the ineffectiveness of integrated care, difficulties in measuring the effectiveness of integrated care interventions using current evaluation frameworks, or both. Integrated care interventions are complex interventions as they are multi-faceted, target patients with complex care needs, try to break the silos between health and social care, and because they have a variety of intended outcomes.[9] These outcomes may not entirely fit within the current frameworks of assessing improvements in quality of life and longevity. It is argued therefore, that it is better to assess the value of integrated care according to the Triple Aim, which includes improved health and well-being, patient experience with care, and reduced costs/improved efficiency. This thesis has shown how Triple Aim outcomes can be assessed in the context of person-centred frail elderly care.

An expanded value-assessment also calls for an expanded evaluation framework and this thesis has shown that Multi-Criteria Decision Analysis (MCDA) offers a suitable framework that can complement (cost-) effectiveness analysis. MCDA can handle a wide set of, sometimes conflicting, outcomes because it uses relative weights which are based on explicit trade-offs between the outcomes, to derive an overall value score.[10] To conduct an MCDA, this thesis has implemented the usual seven steps of MCDA: 1) establish the decision context, 2) identify relevant outcome measures, 3) measure performance of the intervention and comparator on the outcome measures, 4) elicit weights for the outcome measures, 5) create overall value scores, 6) conduct sensitivity analysis, and 7) interpret results together with stakeholders. In our view, this approach can improve the transparency, consistency, accountability, credibility, and acceptability of budget allocation decisions in healthcare, especially at local level.

Research setting

The MCDA performed in this thesis was one of the case studies that was conducted in the European Horizon2020 Project "Sustainable Int**e**grated Care Mode**l**s for Multi-Morbidity: Delivery, **F**inanc**i**ng and P**e**rformance" (SELFIE), which was coordinated by the Health Technology Assessment department of the Erasmus School of Health Policy and Management. SELFIE aimed to contribute to the improvement of person-centred care for persons with multi-morbidity by proposing evidence-based, economically sustainable, integrated care programmes that stimulate cooperation across health and social care and are supported by appropriate financing and payment schemes.

The aim of this thesis was to further advance the economic evaluations of complex interventions in integrated care for persons with multi-morbidity, with a specific focus on frail elderly. This thesis has shed light on the definition and measurement of good health and care as perceived by persons with multi-morbidity and provided an evaluation of a complex integrated care intervention for frail elderly using Multi-Criteria Decision Analysis. Based on the findings from all chapters (see **Table 1)**, it will be discussed what and how this thesis contributes to the care of persons with multi-morbidity, the budget allocation, and the advancement of scientific evaluations in this field.

- **Table 1.** Research objectives and seven steps op MCDA

Objectives	Chapter	Method	Seven steps of MCDA
1. *To unravel the perceptions of persons with multi-morbidity about good health and quality care*	2	Focus groups	Step 2: Identifying outcome measures
	3	Discrete choice experiment	Step 4: Eliciting weights
2. *To empirically evaluate a complex integrated care intervention for frail elderly using Multi-Criteria Decision Analysis*	4	Thick description	Step 1: Describing the decision context
	5	Quasi-experimental study	Step 3: Measuring performance Step 5: Creating an overall value score Step 6: Sensitivity analysis
	6	CEA and MCDA	Step 6: Sensitivity analysis
	7	Deliberation	Step 7: Deliberation

INTERPRETATION OF MAIN FINDINGS

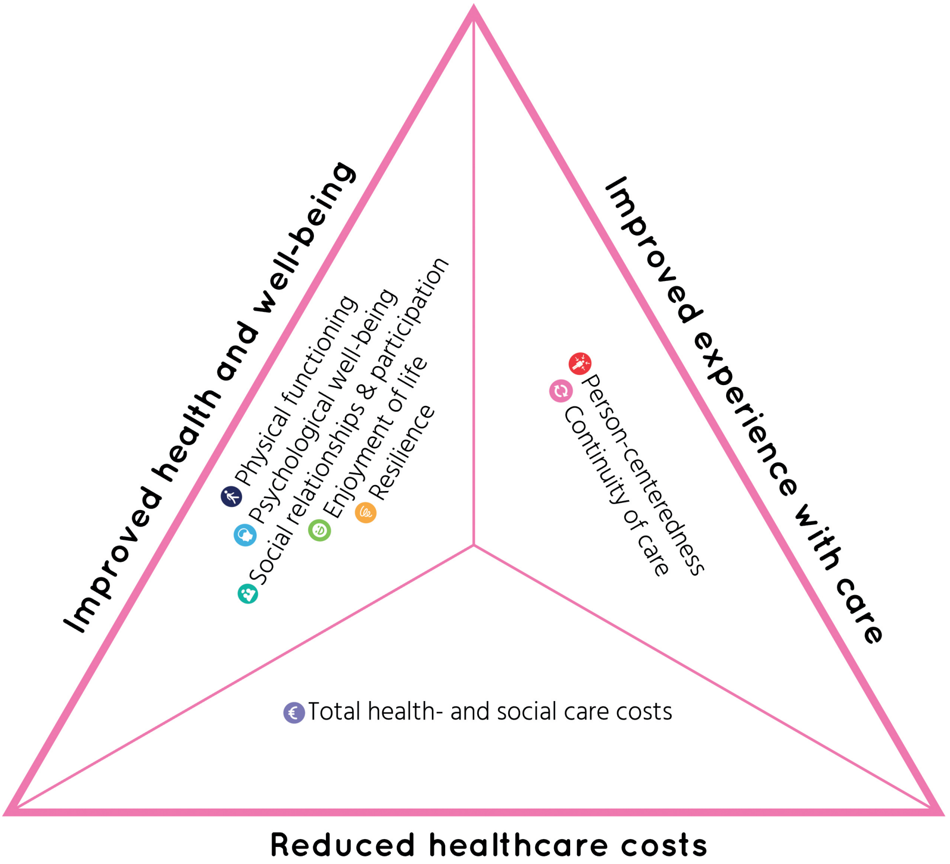
Research objective 1: *To unravel the perceptions of persons with multi-morbidity about good health and quality care*

To address the first research objective, in the first part of this thesis, Chapters 2 and 3 focused on what is (most) important in good health and care for persons with multi-morbidity. To this purpose, focus groups with persons with multi-morbidity were held to identify important outcome measures in health and care (**Chapter 2**) and a core set of outcome measures was then valued in a discrete choice experiment (DCE) to obtain relative importance weights for these outcome measures (**Chapter 3**). Respondents to the DCE not only included persons with multi-morbidity, but also other stakeholder groups involved in integrated care, namely informal caregivers, professionals, payers, and policymakers.

The results of the focus groups reported in **Chapter 2** enabled the incorporation of the "voice" of persons with multi-morbidity in the discussion on meaningful outcomes for empirical evaluations of integrated care. Participants were asked to (1) define what good health and well-being, and a good care process meant to them, and (2) to prioritise the most important outcomes. Results of the focus groups showed that persons with multi-morbidity placed a lot of emphasis on 'enjoyment and pleasure in life', 'freedom and independence', 'psychological well-being', and 'maintaining social relationships and contacts'. When defining a good care process, participants particularly valued well-coordinated and smooth transitions (i.e., continuity of care). This may be inherent to patients facing multiple morbidities as they receive care from multiple care providers and thus will often cross provider-, organisational, and sector boundaries throughout their care trajectory. Also, interactions with care providers were mentioned as important: a respectful treatment, a holistic approach, shared decision-making, and good communication both between care-provider and care-receiver as well as between providers. Aspects related to medical health status and clinical indicators, such as physical functioning, were mentioned to a much lesser extent. Hence, **Chapter 2** concluded that a broad set of outcome measures that entails mental and physical health, well-being, social relationships, and process measures that reflect experience with care delivery should be included in evaluations of integrated care programmes for persons with multi-morbidity.

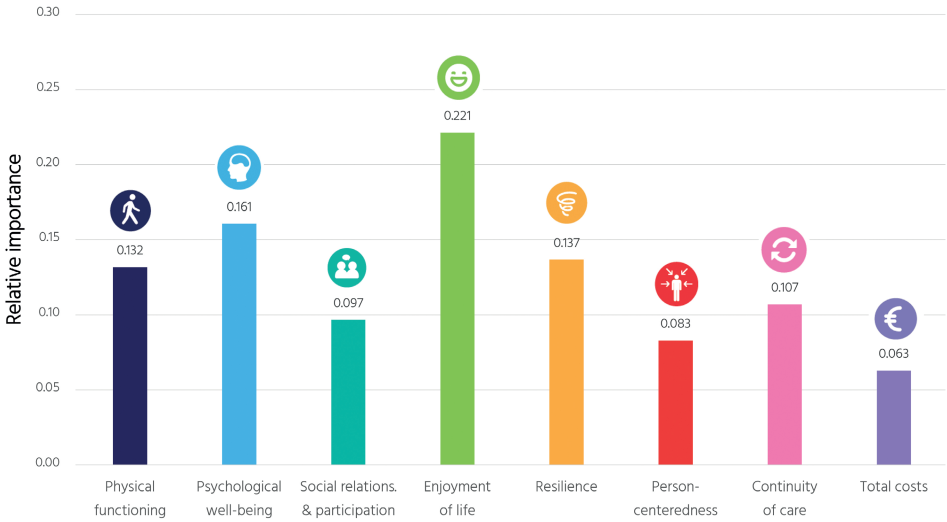
An important step in the MCDA is the identification of outcome measures that will be used throughout the entire process of the MCDA. Results from **Chapter 2** were leading in the selection of the core set of outcome measures, complemented with three other sources that also informed the selection: 1) a literature review carried out for the development of a conceptual framework of integrated care, 2) meetings with national and international stakeholder advisory boards of the SELFIE project that included persons with multi-morbidity, informal caregivers, professionals, payers and policymakers, and 3) a review of the objectives and current outcome measures being used in the 17 SELFIE case studies.[11] The core set of outcome measures that was finally chosen to encompass the Triple Aim of integrated care is presented in **Figure 1**.

● **Figure 1.** SELFIE Core set of outcome measures



Another important step in the MCDA is the elicitation of weights for the selected outcome measures, which was done in **Chapter 3**. The aim of this chapter was twofold, namely, 1) to investigate to which extent outcomes beyond health were valued and 2) to study the heterogeneity of preferences for outcome measures of integrated care among stakeholders involved in integrated care. The DCE consisted of 18 choice tasks, each presenting two hypothetical integrated care programmes that differed in performance on the core set of outcome measures. Respondents were asked to trade-off the outcome measures by choosing which care programme they prefer. The pooled analysis of DCE data showed that the top-3 outcomes with the highest relative importance weight were (1) enjoyment of life, (2) psychological well-being, and (3) resilience (relative weight >0.137 each), see **Figure 2**.

• **Figure 2.** Relative importance weights for the core set of outcomes (all stakeholders combined)



Furthermore, the standard deviations of the preferences for all levels of the outcome measures indicated a wide variation in preferences among respondents, highlighting the heterogeneity in views of stakeholders on what is important in health and care for persons with multi-morbidity. Results of a latent class analysis showed that based on respondents' preferences, four latent classes could be identified with each a different focus on what is most important. The different classes emphasised: experience with care outcomes (class 1, 19.9% of respondents), enjoyment of life (class 2, 39%), physical health (class 3, 18%), whereas class 4 included people without a clear pattern in preferences (24%). Thus, this chapter's results supported results from Chapter 2 and underscored the significance of measuring a variety of outcome measures beyond health – e.g., enjoyment of life and experience with care – to accurately value integrated care.

Research objective 2: *To empirically evaluate a complex integrated care intervention for frail elderly using Multi-Criteria Decision Analysis*

In response to the second research objective, **Chapter 4, 5, and 6** focused on empirically evaluating an integrated care programme using MCDA. For this purpose, the Dutch case study 'Care Chain Frail Elderly' (CCFE) was chosen as an example of an innovative integrated care programme for frail elderly. Hence, this selection enabled us to test the application of MCDA on a very complex population, implying that if it works here it can work in less severe populations as research and evaluation become more challenging with the degree of the complexity of the population. In **Chapter 4**, an elaborate description of the care programme was provided using the method of *thick description*. This led to an in-depth understanding of the decision context and the mechanisms of action of the care programme, which also informed the design of the empirical evaluation carried out in **Chapter 5**. In addition to the MCDA in **Chapter 5, Chapter 6** investigated how MCDA could complement Cost-Effectiveness Analysis (CEA).

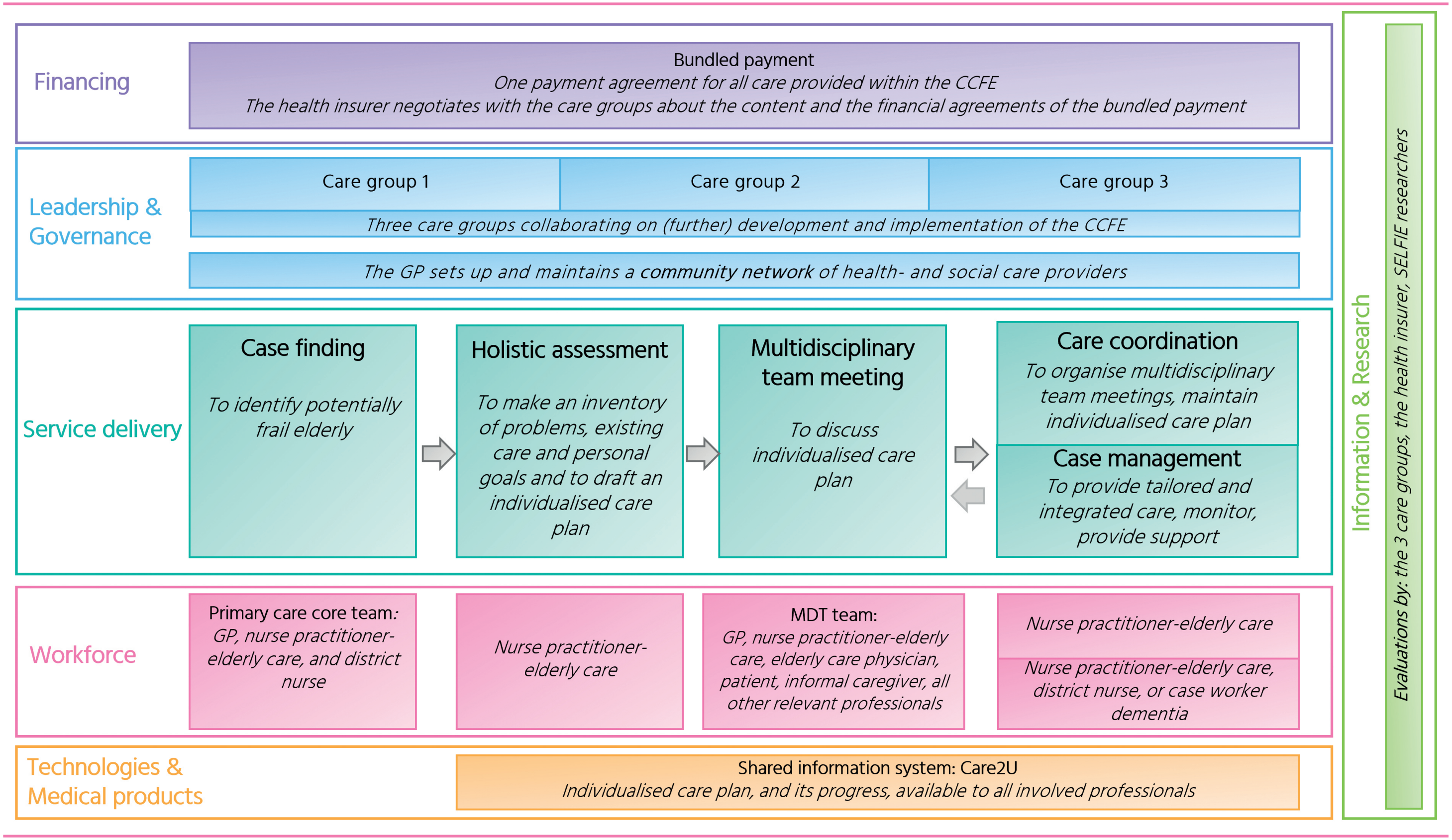
Chapter 4 shed light on the components of the CCFE, their interactions, their implementation in daily practice, and how the programme is perceived by different stakeholders. Thick description covers several levels of depth of analysis and tries to go beyond obvious facts to truly understand what lies beneath the surface and what implicit factors influence components to work. The starting point was a document analysis of a variety of written materials that were already available on the CCFE. Additionally, interviews were conducted with different important stakeholders involved in the CCFE for a deeper understanding of what daily practice in the programme looks like. Results were structured according to the six components of the SELFIE conceptual framework of integrated care: 1) Service delivery, 2) Leadership & governance, 3) Workforce, 4) Financing, 5) Technologies & medical products, 6) Information & research.

The CCFE targets the frailest community-dwelling elderly with complex care needs, identified through a case finding approach that focuses on those most likely to benefit from the programme, thereby refraining from a mere box-ticking exercise when using a fixed checklist. The overall aim of the CCFE was to provide person-centred care coordination and case management to support frail elderly in living at home for as long as possible. Unique features included the presence of frail elderly and their informal caregiver(s) at multidisciplinary team meetings and the innovative way of financing by a bundled payment model. These were considered to be important facilitators for the success of this programme. Other success factors included the holistic assessment of unmet health and social care needs, strong leadership by the care groups and a shared desire and support to continuously improve. The CCFE's innovative components and success factors could be transferred to other settings when providers can take on similar roles and work closely with payers who provide funding that facilitates integration of care. Also, it is recommended to adopt an incremental growth approach and design a shared information system to support smooth collaboration between care providers involved in the programme. A summary of the CCFE according to the six components of the SELFIE conceptual framework of integrated care is given in **Figure 3.**

The empirical evaluation of the CCFE was designed as a quasi-experimental study in which 384 frail elderly persons participated and the performance of the care programme on the core set of outcome measures was measured at three time points (months 0, 6, and 12) by trained interviewers during home-visits (**Chapter 5**). Additionally, patient records on resource utilisation and costs were obtained. Inverse probability weighting was applied to minimise baseline differences between the intervention and control groups. Performance scores were estimated using weighted linear mixed models and subsequently standardised using relative standardisation. In the final MCDA table, all standardised performance scores were weighted according to the perspective of the five stakeholder groups: persons with multi-morbidity, informal caregivers, professionals, payers, and policymakers. Finally, the weighted performance scores were aggregated into overall value scores for both the intervention group and usual care. The results showed that at 6 months follow-up, the CCFE was the preferred way of delivering care to frail elderly. This was mainly driven by enjoyment of life (standardised performance scores 0.729 vs 0.685) and person-centeredness (0.749 vs 0.663), and the high relative importance weight for enjoyment of life across all stakeholder groups. Thus, even though the CCFE does not improve (physical) health as defined in the traditional way, all stakeholders still valued the CCFE positively at 6 months. At 12 months, the MCDA showed little differences between the CCFE and usual care, see **Table 2**. From the informal caregivers' and professionals' perspectives, the CCFE scored higher than usual care, driven by enjoyment of life and person-centeredness. Patients were indifferent between the CCFE and usual care, whereas usual care scored higher than the CCFE according to payers and policymakers. The payers' and policymakers' higher overall value score for usual care could be attributed to the worse performance of the CCFE on physical functioning (0.682 vs 0.731) and costs (€22 816 vs €20 680). Hence, the CCFE may be preferred if enjoyment of life and person-centeredness are considered more important than costs and physical functioning. Vice versa, usual care may be preferred.

As MCDA becomes more relevant than CUA at local level decision-making, the rationale and additional benefits of MCDA over CEA warranted further research. **Chapter 6** aimed to do this by investigating how MCDA could complement CEA to support investment decisions in elderly care at the local level. CEA may not fully capture the value of elderly care innovations, as the aims of these innovations are often geared towards supporting elderly's independence and well-being rather than to improve their health. MCDA may be an appropriate alternative to assessing the value because it allows for the inclusion of a broad set of outcomes and stakeholder perspectives. In this study a side-by-side application of CEA and MCDA, including a comprehensive uncertainty analysis, was performed where the CCFE acted as a case study. Results showed that, according to the CEA and CEAC, the CCFE was not likely (i.e., 36%) to be

- **Figure 3.** Results thick description the CCFE according to the six components of the SELFIE Framework



cost-effective (ICER: € 88,249 from a societal perspective) using the appropriate Dutch willingness-to-pay threshold (i.e., €50,000) that matches the proportional shortfall in the target population. However, the MCDA and associated Conditional Multi-attribute Acceptability Curve (CMAC) demonstrated that the CCFE is potentially equally effective and also affordable as compared with usual care, see **Table 2**. Therefore, the investment decision based on the two approaches could differ. Following results from the MCDA, decision-makers may choose to favour the intervention, because given the uncertainty they could improve enjoyment of life (the most important criterion for all stakeholder groups) and promote person-centred care, which is in line with (national) policy guidance.[12,13]

Final step in MCDA: Deliberation

An integral part of performing an MCDA is deliberation. This seventh step of MCDA was undertaken but it was not reported extensively in the published chapters of this thesis. Results of the focus groups, thick description report, DCE, and MCDA were presented and discussed during the project on multiple occasions, namely, during three national and three international stakeholder workshops with the five stakeholder groups, quarterly meetings with the care groups involved in the CCFE, national and international conferences, and during the final conference where all stakeholder groups were represented. Examples of such meetings were the discussions with representatives from the five stakeholder groups about which outcome measures to include in the study, the meaning of the relative weights assigned to these outcomes in the DCE and their impact on the final results. Moreover, the MCDA results of the CCFE were reflected upon by professionals and payers directly involved in the care programme, for example GPs and representatives from the care groups and the health insurance companies. During these meetings, they deliberated over the performance scores and possible explanations for not finding more distinct differences between the intervention and control group, as some stakeholder had expected. As this approach stimulates mutual understanding among stakeholders and a more transparent, consistent, accountable, credible, and acceptable decision-making process, an even stronger emphasis on deliberation is recommended for future MCDAs.

DISCUSSION OF MCDA IN GENERAL

Broadening the evaluation space

One of the methodological choices we had to make in the SELFIE project, was the selection of outcome measures included in the MCDA (*Step 2*). As the success of integrated care programmes is heavily reliant on the alignment of stakeholder preferences for an agreed set of outcomes, and a discordance in such preferences complicates the decision-making process, it is crucial to include appropriate outcome

- **Table 2.** MCDA table at 12 months: overall value scores for the CCFE and usual care from a societal perspective (Chapter 5 & 6)

Outcome measures	**Standardised performance scores**		**Patients**			**Partners**			**Professionals**			**Payers**			**Policy makers**		
			Weighted score			**Weighted score**			**Weighted score**			**Weighted score**			**Weighted score**		
	CCFE	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**	**Weight**	**CCFE**	**UC**
Physical functioning	0.682	0.731	0.16	0.109	0.117	0.11	0.075	0.080	0.12	0.082	0.088	0.14	0.095	0.102	0.14	0.095	0.102
Psychological well-being	0.701	0.713	0.17	0.119	0.121	0.15	0.105	0.107	0.18	0.126	0.128	0.18	0.126	0.128	0.15	0.105	0.107
Enjoyment of life	0.711	0.703	0.23	0.164	0.162	0.25	0.178	0.176	0.22	0.157	0.155	0.24	0.171	0.169	0.22	0.157	0.155
Social relationships and participation	0.720	0.694	0.08	0.058	0.056	0.09	0.065	0.062	0.11	0.079	0.076	0.10	0.072	0.069	0.10	0.072	0.069
Resilience	0.706	0.709	0.15	0.106	0.106	0.14	0.099	0.099	0.13	0.092	0.092	0.11	0.078	0.078	0.14	0.099	0.099
Person-centeredness	0.744	0.668	0.08	0.060	0.053	0.08	0.060	0.053	0.08	0.060	0.053	0.06	0.045	0.040	0.08	0.060	0.053
Continuity of care	0.719	0.695	0.10	0.072	0.069	0.12	0.086	0.083	0.11	0.079	0.076	0.08	0.058	0.056	0.10	0.072	0.069
Total costs	0.673	0.739	0.03	0.020	0.022	0.06	0.040	0.044	0.06	0.040	0.044	0.08	0.054	0.059	0.07	0.047	0.052
Overall value scores				0.707	0.707		0.708	0.706		0.714	0.713		0.698	0.702		0.706	0.707
95% CI^				0.696 - 0.717	0.697 - 0.717		0.697 - 0.718	0.695 - 0.716		0.697 - 0.717	0.695 - 0.716		0.695 - 0.717	0.697 - 0.719		0.695 - 0.717	0.696 - 0.718
% CCFE>UC				50%			57%			54%			39%			48%	

^ *based on Monte Carlo simulation; Colour scheme ranges from white (lowest score) to green (highest score). CCFE=Care Chain Frail Elderly. UC=Usual care. CI=Confidence Interval.*

measures and to incorporate stakeholder's preferences for these outcomes.[3,14 -16] The importance of the identification and measurement of outcome information is underscored by the ambition of the Dutch ministry of health to have health outcome data available for at least 50% of the total disease burden (which equals 52 conditions, based on Disability Adjusted Life Years (DALYs)) by 2022.[17] In this thesis, the selection of outcomes was largely based on the focus groups with persons with multi-morbidity (Chapter 2), which is consistent with literature recommending including the patient voice in quantitative evaluations.[18-21] The set of outcome measures needed to meet certain requirements to be included in the MCDA, i.e., relevance, completeness, non-redundancy, operationalizability, preferential independence, and sensitivity to intervention-effects (see **General Introduction** of this thesis). However, it is difficult to fully meet these requirements when using outcomes in terms of health and wellbeing. An example of such difficulty is the preference independence condition. To satisfy this assumption, the weight of one outcome measure cannot be dependent on the performance score on another outcome measure. However, interactions between outcomes occur often, meaning that their importance is considered higher or lower than the sum of the importance of the outcomes separately.[22] This is problematic when using a linear additive model. In SELFIE, the core set of outcome measures included both enjoyment of life and psychological well-being, which maybe violated the preferential independence assumption. We still chose to include both outcome measures, as stakeholders perceived them as very important. To meet the assumption as much as possible, we have worded the definition of both outcome measures as differently as possible by stressing the positive connotation in enjoyment of life and the absence of the negative aspects in psychological well-being. Furthermore, we built a constraint in the DCE design, which prohibited the combination of the highest level of enjoyment of life and the lowest level of psychological well-being and vice versa. In future MCDA studies alternative techniques that relax this assumption should be considered.

Moreover, we have shown that MCDA provides a valuable framework to include a broad set of outcome measures beyond the QALY. Although the QALY is a generic measure that eases comparison of cost-effectiveness across interventions and disease areas, it does not include additional elements of value. Our choice to broaden the scope of outcomes measures matches the increased recognition that a standard cost-effectiveness analysis might not be sufficient to reflect all relevant outcomes. This recognition is illustrated by discussions around value-based healthcare (VBHC), augmented cost-effectiveness analysis, the value flower proposed by ISPOR's Special Task Force on US Value Assessment Frameworks, and MCDA.[23 -25] They share the same underlying idea, that current evaluation frameworks do not include all relevant elements of value and should be expanded upon to capture the full value of a treatment or intervention.[26] There has been much debate about these components of value and how to incorporate them in economic evaluation for healthcare decision-making. Policymakers as well as researchers recognise that decision-making incorporates factors that are not considered in the standard cost-effectiveness framework and agree that it may be too restrictive in certain decision contexts. However, including additional elements of value into the evaluation may lead to favouring interventions that do not maximise health. This should not automatically be problematic, as research has shown that individuals are willing to trade-off quality or length of life for these additional elements of value, although the willingness-to-pay for these additional elements appears to be relatively limited.[27,28] However, the question whether society should pay for additional elements that an individual may value should be addressed explicitly. If we think it should, the threshold against which the cost per unit of value is judged needs to be adapted to account for the additional elements of value, because the willingness to pay may change if we (as a society) are not only paying for maximising aggregate health, but also for non-health outcomes.[29,30] Furthermore, interventions that might be displaced when investing in an intervention that improves a broader composite measure of benefit, might produce these additional benefits as well. Hence, the opportunity costs of the intervention changes, when broadening the scope of value.

Besides MCDA, there are other ways to extend the scope of evaluation beyond the QALY. One option that is currently applied to include equity consideration is to weigh QALYs differently to better reflect societal preferences for prioritising relatively younger patients and patients who are relatively more severely ill.[31-35] Equity weights could be based on absolute or proportional shortfall, as is recommended in the UK by the National Institute for Health and Care Excellence (NICE). Recent NICE guidelines propose to use equity weights for therapeutic health technologies; for example if the proportional shortfall is between 0.85-0.95, or the absolute shortfall is between 12 to 18, the QALY weight should be multiplied by 1.2.[36] Another option is to increase the willingness to pay threshold when the severity of disease increases, as we have done in **Chapter 5**, following the Dutch recommendations to adjust the threshold based on the proportional shortfall.[37] A higher threshold for certain categories of drugs or for certain groups (e.g., severely ill) impacts decision-making as interventions that otherwise would not be considered cost-effective may become cost-effective, which might better reflect societal preferences for equitable allocation of resources.[38] A disadvantage of QALY-weighting (option 1) and adjusting the QALY-threshold is that this is only feasible for one of two additional elements that need to be considered in the evaluation.

A third option for expanding the scope of outcomes to consider in an economic evaluation is to conduct a Cost-Benefit Analysis (CBA). This requires that all outcomes are valued in monetary units (often using willingness-to-pay studies) to decide whether the intervention of interest has a positive net benefit over its alternative.[39] An advantage is that the benefits that are valued in willingness-to-pay studies are not restricted to health outcomes. Another framework that can account for a broader concept of value is Social Return on Investment (SROI).[40] It uses monetary values to represent the costs, benefits and disbenefits of an intervention, which are then used to calculate a cost-benefit ratio that presents how many euros are gained in benefits for every euro invested in the intervention.[41] Benefits and disbenefits can pertain to health, social, environmental and economic outcomes. All the benefits (also in terms of cost-savings) are collected and added up. If there are any negative outcomes, they are subtracted, and the result is compared with the costs of the intervention. Hence, all the (dis)benefits are related to the cost of the intervention, whereas in CBA the net monetary benefit based on all costs and monetised benefits is calculated. However, there are many issues in CBA and SROI concerning how health and non-health outcomes can be valued in terms of money and decision-makers may not feel comfortable with monetary value assigned to human lives, quality of life, and outcomes that go beyond health.[39]

A final option that should be mentioned is Cost-Consequence Analysis (CCA). In CCA outcomes are measured in their most appropriate unit but they are not aggregated into a single metric or ratio.[39] Decision-makers are presented with a disaggregated display of all costs and outcomes, which leaves room for the decision-maker to form their own view of the relative importance of the various costs and outcomes of an intervention or treatment. However, it is difficult to take consistent, justifiable, and auditable decisions based on the CCA.

Many disadvantages of abovementioned methods could be overcome by MCDA. For example, in MCDA it is not necessary to place monetary values on outcomes (as is necessary in CBA and SROI) and it provides a method to aggregated both costs and outcomes into an overall value score (which is not done in CCA).

Cost as criterion

The in- or exclusion of cost as one of the criteria in an MCDA is a heavily debated topic.[42,43] When MCDA is used to inform investment or coverage decisions, a decision rule is needed about what constitutes good value for money. An advantage of taking costs into account in the MCDA, is that due to the trade-offs in the weight elicitation procedure, the relative contribution of costs to the overall value score is made explicit. We chose to include costs because the decision context entailed the continuation and wider implementation of a local care programme that was already considered worthwhile to fund by the health insurer. Hence, the question was whether the CCFE in its current form generated sufficient benefits over the comparator (i.e., usual care) to justify the continuation of the investments. Opponents of including costs as a criterion argue that this does not adequately address the opportunity costs of alternative uses of resources.[44] They argue that an MCDA should only include elements of benefit, which then in turn should be compared against the costs to obtain these benefits.[45,46] In that case, the overall value score forms a new composite measure of benefit. The question then becomes "What are the additional cost to generate one additional unit of this new composite benefit score?" Although such an ICER might be an informative indicator of efficiency in and of itself, it should also be compared to a new threshold value that reflects the maximum acceptable ICER for this new composite benefit score. However, such a threshold value is not readily available. It could be derived by using a 'League Table approach' where many interventions are rank ordered according to this new multi-composite ICER, until it exhausts the available budget. In that way, the threshold value would be reflected in the multi-composite ICER of the last intervention possible to fund. Another, more time-consuming option, requires willingness-to-pay studies for these new elements of benefit or estimates of the new composite benefit forgone when interventions are displaced (the opportunity cost). Establishing a threshold must then be repeated, every time that an MCDA creates a new composite benefit score as a result of including context-specific outcome measures. This does not seem feasible, especially not for decision-making at the local level.

Decision-making informed by MCDA

The need for expanding the value assessment in frail-elderly care is evident. However, the discussion above has illustrated how much the aggregation of many different elements of value into a single value metric to support decision-making is debated.[24,47] What if we would not create a single overall value score but only present the element-by-element comparison (similar to a CCA) and leave it to the decision-makers to reach consensus on which intervention provides most value or should be prioritised (deliberation)? The decision-makers would probably not assess all relevant outcomes very systematically, struggle with conflicting information, and have difficulty reaching consensus unless there is a clearly dominant option. They might turn to intuitive or heuristic approaches to simplify the decision-making process, which leads to ad-hoc decisions where other elements of value are only implicitly considered.[48] Moreover, it is likely that the straightforward juxtaposition of the incremental cost-effectiveness ratio and the threshold still receives more attention over qualitative aspects in the deliberation process, at least from decision-makers at the national level.[29,48] As this stands in the way of well-informed and rational decisions, it may diminish the credibility of decision outcomes.[50] In some sectors, like elderly care, healthcare decision-making might just be too complex for a "one-size-fits-all" outcome measure (i.e., cost per QALY) and decision rule (i.e., ICER threshold). Hence, it might be better if decision-makers use a set of complementary methods generating evidence that fits the purpose. This thesis has shown it is worthwhile included MCDA into this set to 1) include a broader assessment of value, and 2) to consider all relevant elements of value systematically, explicitly, and transparently and 3) to do so from multiple perspectives by using stakeholder-specific weights.

It is recognised that the incorporation of multiple weight sets from various viewpoints could also complicate decision-making as a decision-maker needs to decide whose perspective should matter the most when the overall value scores differ between perspectives. We recommend to at least include the patient perspective as they are the end users and their preferences often differ from preferences of other stakeholders.[14,51] Previous research showed that discrepancies in preferences between stakeholder groups could arise due to the influence of patient's own experiences with disease and because they are better informed.[51] Furthermore, we propose to also include the payers' or taxpayers' (i.e., general population) perspective, as the latter are the people who bear the costs associated with healthcare decisions and thus should be part of the decision-making process for the allocation of resources. In our weight elicitation study (*Chapter 3*) we did not observe large differences between stakeholder groups, but this was likely explained by the fact that most stakeholders also identified themselves with additional stakeholder perspectives. If an MCDA would solely be done from the perspective of the general population as is done in CEA for the valuation of QALYs, we would lose the richness of insights that different perspectives can provide.[52]

DISCUSSION OF THE CASE STUDY CCFE

This thesis responds to the international call to expand the use of MCDA models in healthcare decision-making.[24,30] There is a need for greater testing of MCDA models, to learn from these experiences and the issues that arise, and to continuously compare MCDA results with those of standard CEA and similar models. During the seven steps of MCDA, numerous methodological choices were made which warrant close examination and justification. The first methodological decision is the method of MCDA (*Step 1*). Three types are commonly distinguished: value measurement, outranking, or goal programming, which relates to the type of decision the MCDA informs.[53] We chose a value-based method, which aims to assign values to alternatives that reflect preferences regarding the performance of these alternatives. Within value-based methods, the most common approaches are Analytical Hierarchy Process (AHP) and Multi-Attribute Utility Theory (MAUT). AHP is a pairwise comparison method in which the elicitation of weights as well as performance scores are integrated.[54] In MAUT, a single overall value is created by weighted aggregation of standardised performance scores over multiple criteria. We chose Multi-Attribute Value Theory (MAVT), which is a simplification of MAUT, as MAVT does not seek to model the decision-maker's attitude to risk.[55] This makes the weight elicitation choice tasks easier to comprehend. Furthermore, an advantage of MAUT over AHP is that the relative importance weights could be determined in a separate trajectory, which enhances the reusability of the MCDA framework. We did not opt for AHP as this comes with its own challenges, such as rank reversal – i.e., changes in the relative ranking of the original alternatives, when a new alternative is introduced.[56]

Challenges to obtain empirical performance scores using causal inference

Quasi-experimental study design

Insights into the care programme demonstrated that the CCFE is a comprehensive, time-intensive, and costly intervention, only made available for the frailest community-dwelling older persons. This was reflected in the high expectations of the care program's value, especially in terms of the well-being and experience with care outcome measures. To measure the performance of the care programme (Step 3), a quasi-experimental study design was setup as randomisation was not possible because the programme was already being implemented in daily practice and it was considered unethical to withhold treatment from patients. Therefore, a control group was formed with frail elderly selected at general practitioners that did not provide the CCFE. These general practitioners belonged to one of the three care groups that offered the CCFE but had not started providing the intervention. Despite the high expectations of the care programme, 12-month results were not convincing which evoked discussion about the evidence with the care groups and during the stakeholder workshops. Besides the possibility that initial effects might be hard to maintain over a longer term, there are several other potential explanations that merit attention. One possible explanation is that even though we compared the intervention group to 'usual care', it is difficult to demarcate usual care from intervention care. For example, case management – which is an essential element of the CCFE – could also be part of usual care, especially in case of dementia, and an individual care plan is also often part of the chronic care programmes for diabetes, COPD and (cardio)vascular risk management. In the control group, participation in the two largest chronic care programmes amounts to 42% in (cardio)vascular risk management and 26% in diabetes. This reflects that in the Netherlands there is already a strong primary care sector, which makes it harder to demonstrate the impact of proactive integrated care compared with usual care.[57] This was also stressed by other studies comparing integrated care with usual care in the Dutch primary care setting.[58] Another possible explanation for the fact that the impact seemed to be less than anticipated is an overall shift in expectations about how good quality of life and the process of care delivery and support should be at old age.[59]

Difficult to reach target group

The target population of the CCFE was a difficult to reach group, namely, the frailest elderly persons still living at home. As previous research confirms, it is a challenge to collect data among this group and it is important not to underestimate the difficulty of this task, and to ensure that the data collection period is sufficient to recruit adequate numbers.[60-62] The close collaboration with the care groups, which included care providers directly involved with the target group, was beneficial for the set-up of the study design, which was essentially co-created. The care groups emphasised that the frail elderly would not be able to independently fill in the survey or participate in telephone interviews. Moreover, they pointed out that informal caregivers could also not be burdened with the additional task of assisting in the data collection. Hence, we chose to administer the questionnaire during home visits, which ensured that many vulnerable elderly persons could still participate. It also removed the concerns that informal caregivers would interfere and answer on behalf of the elderly, although sometimes informal caregivers were present during the interviews. However, besides all the efforts to reach the target group, it was still not possible to include elderly with more than mild cognitive impairments, for obvious reasons. This may affect the external validity of the study, as even among the community-dwelling frail elderly a considerable proportion may suffer from dementia or other cognitive impairments.

Choice of weight-elicitation method

A further choice relates to the weight elicitation method (*Step 4*). Commonly used weight-elicitation methods are DCE, AHP, swing-weighting, direct ranking, and point allocation.[63] These different techniques differ in the level of cognitive challenge they pose, which played an important role as an important stakeholder group were patients who may not be familiar with the tasks they were asked to complete.[53] We chose to perform DCEs as it is a theoretically well-founded method to elicit stated preferences.[64-67] Important assets of this method are that (1) in DCEs the entire potential range of performance on an outcome measure (i.e., attribute) is considered (lowest level to highest level) and (2) in DCEs stakeholders are forced to trade criteria off against one-another, as opposed to merely rating a single criterion.[68] To ensure that patients were able to comprehend the questionnaire, we pilot-tested the DCE among patients, and included practice questions at the start of the questionnaire. Throughout the survey, participants were able to request additional information regarding the definitions of the outcome measures they were asked to trade-off. A DCE generally does not involve interaction between participants, which hampers discussion between stakeholders. However, in SELFIE we discussed results from the DCE in (inter)national stakeholder workshop to discuss any concerns or striking results. The generated weight sets are of great value to the reusability of the MCDA, for example by other researchers evaluating integrated care programmes using conceptually similar outcome measures as in our study. There is already one study that mapped their outcome measures to the SELFIE core set of outcomes and used the SELFIE weight sets to carry out an MCDA.[69] To stimulate the reusability, we have developed an online MCDA tool with detailed instructions that researchers could use free of charge.[70] It includes the weight sets from all stakeholder groups in all participating SELFIE countries, and an averaged weight set across all countries.

Patient engagement

Aside from the methodological choices, the experience gained from this thesis highlights several advantages of using MCDA in the current context. Most importantly, throughout the entire process of the MCDA, the patient voice was actively involved. Patients were the main source of information for the selection of the core set of outcomes, patients participating in the care programme were interviewed to learn how they experienced the care programme, and patients were included as one of the five stakeholder groups. The MCDA was first performed from their perspective, applying their relative weights for the outcome measures. Patients were invited to each stakeholder workshop and were an active voice during the deliberation upon the results.

Implementation and upscaling

In the last phase of the SELFIE project we focused on drivers of successful implementation of integrated care for multi-morbidity, and empirically identified 10 mechanisms that successful implementers had applied.[71] To summarise, these successful implementation mechanisms advise to 1) engage the stakeholders in alignment work (e.g., aligning components in an individualised care plan), 2) adopt an incremental rather than a disruptive growth model, 3) balance between flexibility and formal structures of integration, 4) apply collaborative governance (by engaging all stakeholders), 5) distribute leadership throughout all levels of the system, 6) build a multidisciplinary team culture, 7) develop new roles and competencies for integrated care, 8) secure long-term funding and adopt innovative payment that overcome fragmentation, 9) implement ICT to support collaboration and communication, and 10) create feedback loops & continuous monitoring. Two of the most distinctive mechanisms applied in the implementation of the CCFE pertain to developing new roles, as the CCFE invites frail elderly to participate in the multidisciplinary team meetings and thus expect them to take on a new role as active member in the multidisciplinary care team. This also required that care providers strengthened or learned new competencies such as shared decision-making. Secondly, the care groups put a lot of effort in securing long-term funding by adopting an innovative payment in the form of a newly developed bundled payment that transcends the existing bundled payments. Funding transitioned from short-term financing on a project basis, to ‘Elderly care modules’ as extra financing to stimulate multidisciplinary elderly care, to annual negotiations about the continuation and content of the bundled payment for frail elderly, to biannual negotiations with the health insurer.[72] The involvement of the insurer was seen as an asset as it provided promise for the financial sustainability of the CCFE. This ensured continuity and opportunities to further develop and improve the care programme. Also other mechanisms can be recognised, such as alignment work (by drafting individualised care plans), adopting an incremental growth model starting in 2011 (e.g., by gradually expanding from one care group to the other care groups, and by increasing the number of GP-practices that offer the CCFE), building a multidisciplinary team culture, and implementing ICT to support collaboration and communication (by using a shared information system across all involved care providers).

FUTURE DIRECTIONS

In policy

Currently, cost-effectiveness analyses are used to generate evidence about the value and the costs of treatments or interventions to aid reimbursement decisions. Next to economic evaluations, these decisions are often informed by additional information, for example on the burden of disease, vulnerability of the target population, and the budget impact. We propose to use a broader measurement of value for integrated care interventions for persons with multi-morbidity like frail elderly, including outcomes that span the Triple Aim, namely: physical functioning, psychological well-being, enjoyment of life, social relationships & participation, resilience, person-centeredness, continuity of care, and costs. Many of those outcomes beyond health are valued as very important by persons with multi-morbidity themselves.

Furthermore, we argue that MCDA should be used in addition rather than as a replacement of conventional cost-effectiveness analysis. Where CEA could provide relevant information to compare various diseases areas and treatment options – relevant on the national level – MCDA provides additional information beyond the – in some circumstances – too narrow definition of value used in CEA. This could especially be useful for local level decision-making in elderly and social care, where interventions tend to focus on improving outcomes beyond health and longevity. Such interventions often aim to improve well-being, maintain independence, and increase satisfaction with the care process. Thus, to adequately compare or rank multifaceted elderly care programmes, broader outcome measures should be considered.

In practice

Integrating care is an ongoing process that continuously deserves attention to develop, improve, and implement innovative care programmes that focus on improving relevant elements of value. Furthermore, it should be encouraged to participate in research to enable comparative analysis of different approaches towards integrated care for frail elderly. To generate evidence, future programmes should incorporate an ongoing monitoring process that includes a clear set of outcome measures, validated instruments to measure the selected outcome measures, and a suitable evaluation framework that combines these data and aids nuanced, explicit, and transparent decision-making. The SELFIE MCDA core set of outcomes is specifically appropriate for integrated care for persons with multi-morbidity and therefore advised for this setting. To ensure feasibility of data collection, the extensive SELFIE questionnaire used (in a research setting) to measure performance for the SELFIE outcome measures may need to be transformed into a minimal dataset to measure the performance of alternatives using a small set of indicators implemented in routinely collected data. Furthermore, such programmes could make use of the SELFIE weight sets. Following this direction, the intervention could be monitored and continuously adapted based on results. This is in accordance with the aforementioned mechanism of successful implementation to create feedback loops and continuous monitoring. Accordingly, an MCDA framework would improve the acceptability of innovations by (1) involving the patients as they can get information on outcomes they can comprehend, such as enjoyment of life and person-centeredness; (2) engaging healthcare and social care professionals, as they see the reflection of their work on intermediate outcomes such as person centeredness which helps to stay motivated; and (3) giving insights to managers/commissioners/payers, as they can monitor the performance of interventions on short- and medium term outcomes while keeping services within budgets.

In research

Researchers evaluating integrated care programmes are advised to include a broad set of outcome measures that cover the Triple Aim of integrated care to fully capture the value of such interventions. Future studies measuring similar outcomes as the SELFIE core set of outcome measures are encouraged to use the MCDA tool. The MCDA tool is very accommodating to other study designs and methods, as it could also be used when not all outcome measures are measured or when other instruments are used to collect a conceptually similar outcome.[69] However, this is not ideal as the relative importance weights must be rescaled to adjust for the omitted outcomes, and it is not clear whether the omission of outcomes would have affected the trade-offs between the outcomes. Furthermore, it could also be used with weight sets from other countries, or with average weights from all countries that participated in the SELFIE project. MCDA could also be used for other disease areas and interventions as its benefits also apply outside of elderly care. This requires a new selection of outcome measures and corresponding weight sets. An important topic on the future research agenda should be how results from MCDA could be compared across disease areas and treatments.

Finally, the large number of issues that arise with expanding value assessments and the inclusion of additional elements of value in healthcare decision-making, as described above, points to the overall importance of further researching how to incorporate value in healthcare decision-making.

CONCLUDING REMARKS

The most important contribution of this thesis is the development and application of an MCDA framework to evaluate integrated care using primary and secondary data, as well as a range of qualitative and quantitative methods. We have shown that persons with multi-morbidity value outcomes beyond health, when asked about good health and healthcare. We created a core set of outcomes that we recommend being included in future economic evaluations of integrated care for multi-morbidity: physical functioning, psychological well-being, enjoyment of life, social relationships & participation, resilience, person-centeredness, continuity of care, and total health- and social care costs. Subsequently, in a weight-elicitation study we found heterogeneity in preferences confirming that it is crucial to include a variety of relevant outcome measures that match the aims of the integrated care programme. Therefore, we advocate that a broad set of outcomes that entail health, well-being and experience of care should be included when evaluating integrated care for persons with multi-morbidity, and important next steps include moving towards harmonising evaluation frameworks. The second part of this thesis demonstrated that carrying out the seven steps of MCDA of an integrated care programme for frail elderly provides a deep understanding of the workings of the intervention and its decision context (when applying a thick description approach), and a broader insight into the value of the intervention. We demonstrated that uncertainty in MCDA could be addressed with probabilistic sensitivity analysis and the CMAC. MCDA complements CEA with information beyond health and longevity, which can lead to different statements about the added value of a care programme as we have shown.

All these contributions could move MCDA a step further in informing healthcare decision-makers to achieve the multiple objectives of the health system, which include besides longevity and health-related quality of life, improving well-being, access to care and protection of the most vulnerable, patient experience with care, and efficiency. The comprehensive view on the value of a care programme that MCDA gives, from the perspective of patients, informal caregivers, professionals, payers, and policymakers, aids decision-making in elderly and social care—where interventions are generally pointed at improving broader outcomes—because decisions regarding reimbursement or scaling-up become more transparent, explicit, and nuanced. This, in turn, fosters the acceptability and implementability of integrated elderly care innovations.

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Chapter

8

Appendices

8

Summary

SUMMARY

The rapidly ageing population in the Netherlands urges health system transformations towards integrated care to improve effectiveness, efficiency and maintain affordability. Major healthcare reforms have incentivised older persons to live independently for as long as possible, with support in their own environment. However, ageing is associated with multi-morbidity and most frequently with frailty. Hence, many older people will receive care and services from multiple organisations and professionals across the health- and social care spectrum. Although these care providers aim to support frail elderly, there is room to improve their collaboration and reduce the fragmentation and inefficiency of the care provision..

To overcome fragmentation and its negative consequences, novel programmes have been developed that integrate care by focusing on delivering person-centred care in a continuous process. In these programmes, older people are approached holistically, and care providers improve the communication with one-another to tailor the services to the needs, capabilities, and preferences, and smoothen transitions between services and reduce overlapping treatments. These integrated care programmes require assessment of their effectiveness and cost-effectiveness to justify payment from public resources. Yet, current (economic) evaluation frameworks might not provide sufficient insight into the broad range of outcomes that such integrated care programmes aim to improve. Currently, cost-utility analysis – which is the gold standard in economic evaluations that aim to inform reimbursement decisions – focus solely on longevity and health-related quality of life. Multi-Criteria Decision Analysis (MCDA) could complement cost-utility analysis, by 1) incorporating a broader set of outcome measures covering the Triple Aim of integrated care, and 2) adopting multiple-stakeholder perspectives on the importance of the outcome measures. The results of MCDA are overall value scores (for both an intervention and usual care) that give insight into the added value of an intervention from multiple perspectives. Hence, MCDA has the potential to improve transparency, consistency, accountability, credibility, and acceptability of decision-making.

An MCDA is commonly conducted in seven steps, which were addressed in the several chapters of this thesis. They include: 1) establishing the decision context, 2) identifying relevant outcome measures, 3) measuring performance of the intervention and comparator on the outcome measures, 4) eliciting weights for the outcome measures, 5) creating overall value scores, 6) performing sensitivity analysis, and 7) interpretation of results.

Research setting

The research took place in the European Horizon2020 Project "Sustainable Integrated Care Models for Multi-Morbidity: Delivery, Financing and Performance" (SELFIE). SELFIE aimed to contribute to the improvement of person-centred care for persons with multi-morbidity by proposing evidence-based, economically sustainable, integrated care programmes that stimulate cooperation across health and social care and are supported by appropriate financing and payment schemes.

Research aims

As the scarce evidence of the (cost-)effectiveness of person-centred integrated care for older people is often of low methodological quality and provides a mixed picture, the question was raised how to design adequate (economic) evaluations of complex integrated care interventions for this challenging target population. Therefore, this thesis aimed to further advance (economic) evaluations of integrated care by addressing the following research aims:

1. *To unravel the perceptions of persons with multi-morbidity about good health and quality care*
2. *To empirically evaluate a complex integrated care intervention for frail elderly using Multi-Criteria Decision Analysis*

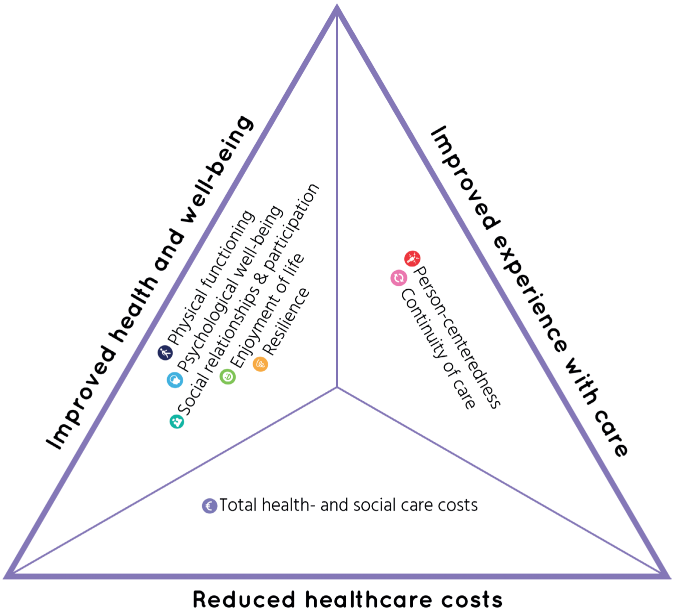
Part 1. Unravelling the perceptions of persons with multi-morbidity about good health and quality care

The first part of this thesis explored what comprehends good health and care for persons with multi-morbidity, from the perspective of patients themselves, and from the viewpoint of informal caregivers, professionals, payers, and policy makers.

Chapter 2 presents the results of focus groups held with persons with multi-morbidity in eight European countries: Austria, Croatia, Germany, Hungary, the Netherlands, Norway, Spain, and the United Kingdom. This qualitative approach ensured that perspectives of persons with multi-morbidity are incorporated in the discussion on what meaningful outcomes for empirical evaluations of integrated care are. Participants were asked to (1) define what good health and well-being, and a good care process meant to them, and (2) to prioritise the most important outcome measures. Participants across all countries placed a lot of emphasis on 'enjoyment and pleasure in life', 'freedom and independence', 'psychological well-being', and 'maintaining social relationships and contacts'. When defining a good care process, participants particularly valued well-coordinated and smooth transitions (i.e., continuity of care). This might be explained by the fact that patients facing multi-morbidity have multiple care providers and thus will often cross provider-, organisational, and sector boundaries throughout their care trajectory. Also, interactions with care providers were mentioned as important: a respectful treatment, a holistic approach, shared decision-making, and good communication both between provider and participant as well as between providers. Interestingly, aspects related to medical health status and clinical indicators, such as physical functioning, were mentioned to a much lesser extent. The study concluded that a broad set of outcome measures that entail mental and physical health, well-being, social relationships, and process measures that reflect experience with care delivery should be included in evaluations of integrated care programmes for persons with multimorbidity.

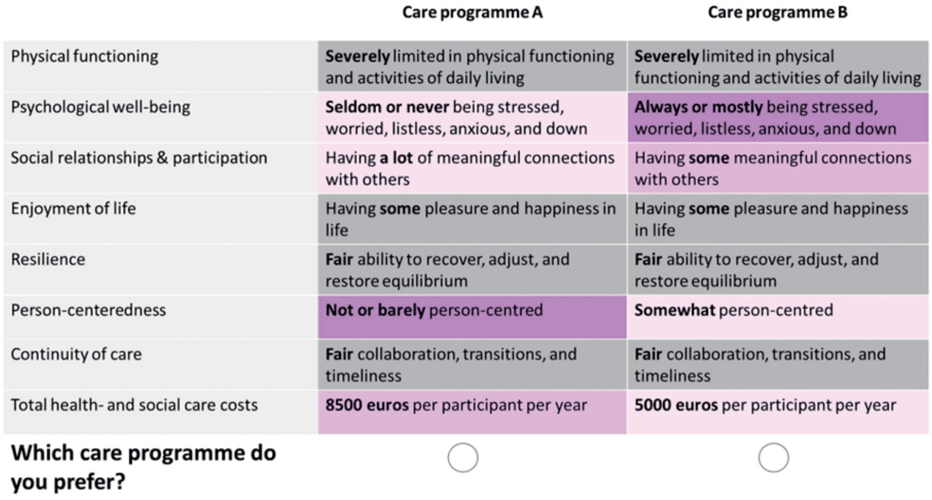
Chapter 2 fed into the design of the next chapters as the results formed the basis for the 'core set of outcome measures' established in the SELFIE project, which played a central role throughout this thesis. Although the focus groups laid the foundation for the formation of the core set, three other sources also informed this process: 1) a literature review carried out for the development of a conceptual framework of integrated care, 2) meetings with national and international stakeholder advisory boards of the SELFIE project that included patients, informal caregivers, professionals, payers and policy makers, and 3) a review of the aims and current outcome measures being used in the 17 SELFIE case studies (among which the Care Chain Frail Elderly). These four sources culminated in the core set of outcome measures that encompasses the Triple Aim of integrated care, see **Figure 1**.

- **Figure 1.** SELFIE Core set of outcome measures



Chapter 3 describes the results of a pooled analysis of the discrete choice experiments (DCEs) conducted among patients, informal caregivers, professionals, payer, and policy makers. The aim of this chapter was twofold, namely, 1) to investigate to which extent outcome measures beyond health are valued, and 2) to study the heterogeneity of preferences for outcome measures of integrated care among stakeholders involved in integrated care. In the DCE, respondents were presented with two hypothetical integrated care programmes that differed in performance on the core set of outcome measures. They were asked to choose which care programme they prefer, hereby making trade-offs between multiple outcome measures of an integrated care programmes. The relative importance of each outcome measure was based on the coefficient of its best level (level 3) divided by the sum of all best attribute levels. An example of such a so-called 'DCE choice task' is presented in **Figure 2**.

- **Figure 2.** Example of a DCE choice task



	Care programme A	Care programme B
Physical functioning	**Severely** limited in physical functioning and activities of daily living	**Severely** limited in physical functioning and activities of daily living
Psychological well-being	**Seldom or never** being stressed, worried, listless, anxious, and down	**Always or mostly** being stressed, worried, listless, anxious, and down
Social relationships & participation	Having **a lot** of meaningful connections with others	Having **some** meaningful connections with others
Enjoyment of life	Having **some** pleasure and happiness in life	Having **some** pleasure and happiness in life
Resilience	**Fair** ability to recover, adjust, and restore equilibrium	**Fair** ability to recover, adjust, and restore equilibrium
Person-centeredness	**Not or barely** person-centred	**Somewhat** person-centred
Continuity of care	**Fair** collaboration, transitions, and timeliness	**Fair** collaboration, transitions, and timeliness
Total health- and social care costs	**8500 euros** per participant per year	**5000 euros** per participant per year
Which care programme do you prefer?	○	○

The pooled analysis of DCE data showed that the top-3 outcomes with the highest relative importance weight are (1) enjoyment of life, (2) psychological well-being, and (3) resilience. Total health- and social care costs was considered the least important outcome of an integrated care programme. **Figure 3** presents the relative importance weights for the core set of outcomes. The standard deviations of all outcome levels indicated a wide variation in preferences among respondents, which triggered the question whether there were subgroups to be detected with similar preferences. Therefore, latent class analysis was used to identify subgroups of respondents (i.e., latent classes) that share similar preferences for certain outcome measures based on their observed choice data. Subsequently, we explored the respondents' characteristics of each subgroup.

● **Figure 3.** Relative importance weights for the core set of outcomes

The latent class analysis identified four classes that each put emphasis on specific outcomes: (1) assigns a relatively higher weight to experience with care, (2) assigns a relatively higher weight to enjoyment of life and psychological well-being, (3) assigns a relatively higher weight to physical and mental health, (4) 'indifferent'. Differences among the classes' preferences were only weakly related to whether respondents were patients, informal caregivers, professionals, payers, or policymakers, as many stakeholders indicated they take on multiple roles. This heterogeneity in preferences underlines the importance of taking into account a wide range of outcome measures, including well-being and experience with care outcomes, when setting up and evaluating integrated care programmes.

Part 2. Evaluation of a complex integrated care intervention for frail elderly

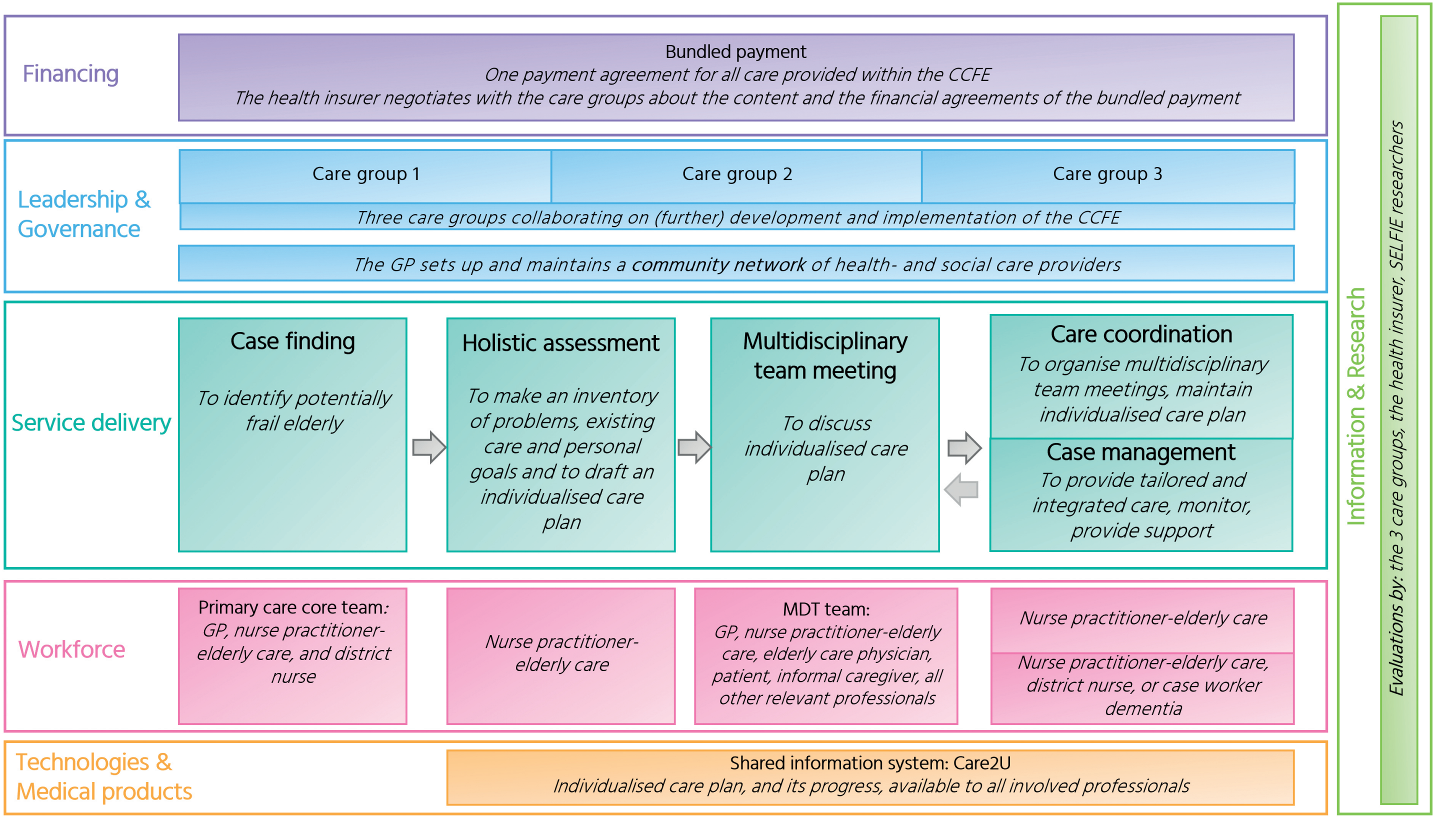
The second part of this thesis focuses on empirically evaluating an integrated care programme, taking the Care Chain Frail Elderly (CCFE) as a case study. The evaluation entailed gaining a deep understanding of the CCFE and the decision context, measuring the performance of the CCFE and its comparator (usual care) on the core set of outcomes, and combining the performance with the relative importance weights, hereby performing an MCDA.

In **Chapter 4** the CCFE is systematically described and analysed using a thick description approach that combines document analysis and qualitative interviews to gain an in-depth understanding of the programme. The overall goal was to identify the barriers and facilitators of implementation, and to highlight innovative elements of the CCFE, so that these insights may inform the implementation process and design of evaluation studies of similar programmes in the future. The thick description method was chosen as it covers several layers of depth of analyses and aims to reach beyond the descriptive nature of document analysis. It allows for a thorough investigation of implicit social practices while also considering the specific context of the intervention. The interviews with key stakeholders (e.g., initiators, patient, informal caregiver, GP, nurse practitioner) complemented the written documents by providing a deeper understanding of what constituted the programme below its surface and when put into practice. The results were structured according to the conceptual framework of integrated care developed in the SELFIE project, to describe, develop, and implement integrated care programmes. The framework consists of the following six components that were addressed at the macro-, meso- and micro level, while putting the individual at the centre: 1) Service delivery, 2) Leadership & governance, 3) Workforce, 4) Financing, 5) Technologies & medical products, 6) Information & research.

The CCFE targets community-dwelling frail elderly with complex care needs and aims to provide person-centred care coordination and case management to support frail elderly in living at home for as long as possible. The programme consists of several key elements that are common in frail elderly programmes, e.g., a holis¬tic assessment of unmet health and social care needs, individualised care planning, multidisciplinary care, care coordination, and case management. However, the CCFE distinguishes itself from other initiatives by targeting the frailest GP-population, direct engagement of the patient and informal caregiver in the multidisciplinary team meetings, and the innovative financing by means of a bundled payment. A summary of the CCFE and its most distinctive elements is given in **Figure 4**. We concluded that results could inform others developing similar programmes as the CCFE's innovative components and critical success factors are likely to be transferable to other settings.

Chapter 5 presents the results from the Multi-Criteria Decision Analysis of the CCFE. To assess the performance on the core set of outcomes of the CCFE and usual care, data were collected among 384 frail older persons at 0, 6 and 12 months by trained interviewers during home-visits.

Figure 4. Results thick description of the CCFE according to the six components of the SELFIE Framework



The 6-month results showed that the CCFE performed better than usual care on person-centeredness. However, physical functioning declined in both groups but even further in the intervention group. At 12 months, the CCFE performed better than usual care on person-centeredness, but differences across all outcomes were small, see **Figure 5**. Subsequently, the performance scores were multiplied with the relative importance weights from the five stakeholder perspectives. The MCDA results indicated that the CCFE is the preferred way of delivering care to frail elderly at 6 months. At 12 months, MCDA results showed little differences between the CCFE and usual care from the perspective of patients, informal caregivers, and professionals, while payers and policy makers seemed to prefer usual care. Hence, the CCFE may be preferred if enjoyment of life and person-centeredness are considered more important than costs and physical functioning.

- **Figure 5.** Standardised performance scores of the CCFE and usual care at 12 months

Note: Left = the CCFE, right = usual care. Results are standardised, meaning they are all on the 0-1 scale and a higher score indicates a better performance (for costs a better performance means lower costs)

Furthermore, we advocate a wider use of MCDA to evaluate multi-faceted integrated care that has a variety of intended outcomes that reach beyond quality of life and life extension. This approach could then provide useful information to enable a transparent, consistent, accountable, credible, and acceptable decision-making process of value-based care.

Chapter 6 further explores the applicability of MCDA by investigating how MCDA could complement Cost-Effectiveness Analysis (CEA) to support local level decision-making. Additionally, it introduces the Conditional Multi-attribute Acceptability Curve (CMAC), which represents the probability that the CCFE has the highest overall values score (i.e., is the preferred alternative) while the budget (given a certain target group size) remains below a set threshold for the available budget. Results of the CEA, using a willingness-to-pay threshold of 50,000 euros per QALY (Quality-Adjusted Life Year), showed that the CCFE was not likely to be cost-effective. However, from an MCDA perspective, the CCFE demonstrated that it is potentially equally effective and affordable as usual care, while also improving enjoyment of life and person-centeredness. Therefore, the investment decision based on the two approaches could differ when policy makers opt to fund innovations that provide more value as assessed by a broad set of outcome measures including enjoyment of life and person-centeredness. Thus, MCDA complements CEA in that it provides decision-makers with a more comprehensive depiction of effectiveness of an integrated care programme, for example along the lines of the Triple Aim of integrated care. Whether to apply CEA or MCDA to inform decisions, depends on the decision context. Conventional CEA might be best suited for decision-making on the national level regarding the choice of treatments that should be added to the benefit package as it allows for more direct comparisons between disease areas and treatments. However, a CEA may not fully capture the impact of integrated elderly care interventions that often combine health and social care and aim to improve outcomes beyond health. MCDA could then complement information on the value of integrated care interventions, which could be especially useful in local-level decision-making. These findings are relevant, as it could foster the acceptability and implementability of innovations in elderly care.

Finally, the main findings of this dissertation were interpreted and discussed. The final chapter reflects on the theoretical and methodological considerations and sheds light on the implications for policy, practice, and research.


8

Nederlandse samenvatting

SAMENVATTING

De komende jaren neemt het aantal ouderen in Nederland rap toe. Een transformatie van het zorgsysteem naar integrale zorg kan bijdragen aan het efficiënter organiseren en betaalbaar houden van de zorg. Door de ingrijpende hervorming van de ouderenzorg in Nederland worden ouderen gestimuleerd om zo lang mogelijk thuis te blijven wonen, met ondersteuning dicht bij huis. Echter, ouder worden gaat vaak gepaard met multimorbiditeit en kwetsbaarheid en daardoor ontvangen ouderen zorg en ondersteuning van verschillende organisaties en zorgverleners. Hoewel deze zorgverleners het doel hebben om de oudere zo goed mogelijk te ondersteunen, is er veel ruimte om de samenwerking te verbeteren en de fragmentatie en inefficiëntie in de zorg te verminderen.

Om fragmentatie en de bijbehorende negatieve gevolgen te voorkomen, zijn er nieuwe integrale zorgprogramma's ontwikkeld die zich richten op een persoonsgericht zorgproces en op continuïteit van zorg. Ouderen worden benaderd op een holistische wijze, en zorgverleners communiceren regelmatig met elkaar om de zorg en ondersteuning af te stemmen op de behoeften, mogelijkheden en voorkeuren van de ouderen, om overgangen tussen zorg te versoepelen en om overlap in de zorg te verminderen. Om vergoeding van deze integrale zorgprogramma's uit publieke middelen te rechtvaardigen, moet de effectiviteit en kosteneffectiviteit van de zorgprogramma's getoetst worden. De huidige (economische) evaluatiemethoden zijn vaak niet geschikt om inzicht te geven in de diverse uitkomsten die zulke integrale zorgprogramma's beogen te verbeteren. In kosten-utiliteitsanalyses – die worden gezien als de gouden standaard in economische evaluaties om vergoedingsbeslissingen te onderbouwen – wordt alleen gekeken naar de uitkomstmaten levensduur en gezondheidsgerelateerde kwaliteit van leven. *Multi-Criteria Decision Analysis* (MCDA) zou kosten-utiliteitsanalyses op twee manieren kunnen aanvullen, namelijk; door 1) het meenemen van een bredere set van uitkomstmaten die de *'Triple Aim'* (*NL = Drie Doelen*) van integrale zorg omvat, en 2) het meenemen van r het relatieve belang van de uitkomstmaten vanuit de optiek van verschillende groepen stakeholders. Een MCDA resulteert in *'overall value scores'* (*NL = algehele waardescore*) (voor zowel de interventie- als de controlegroep) die inzicht geven in de waarde van een interventie (of gebruikelijke zorg), en vanuit welk (stakeholder-) perspectief deze waarde wordt ervaren. Daarom heeft MCDA het potentieel om de transparantie, consistentie, aansprakelijkheid, geloofwaardigheid en aanvaardbaarheid van beslissingen te verbeteren.

MCDA bestaat vaak uit zeven stappen, die in de verschillende hoofdstukken van dit proefschrift aan de orde kwamen: 1) de beslissingscontext vaststellen, 2) relevante uitkomstmaten identificeren, 3) de prestatie van de interventie- en controlegroep op de uitkomstmaten meten, 4) relatieve gewichten bepalen voor de uitkomstmaten, 5) het creëren van de *overall value scores*, 6) sensitiviteitsanalyses uitvoeren en 7) interpretatie van de resultaten.

Onderzoekssetting

Het onderzoek was onderdeel van het Europese Horizon2020 Project "Sustainable Integrated Care Models for Multi-Morbidity: Delivery, Financing and Performance" (SELFIE). SELFIE beoogde een bijdrage te leveren aan de verbetering van persoonsgerichte zorg voor mensen met multimorbiditeit door empirisch onderbouwde, economisch duurzame, integrale zorgprogramma's voor te stellen die de samenwerking tussen gezondheids- en sociale zorg stimuleren en worden ondersteund door passende financierings- en bekostigingssystemen.

Onderzoeksdoelen

Het relatief weinige bewijs voor de (kosten-) effectiviteit van persoonsgerichte integrale zorg voor ouderen is vaak van methodologisch slechte kwaliteit en geeft een inconsistent beeld van de uitkomsten. Dit riep de vraag op hoe evaluaties van complexe integrale zorginterventies voor deze lastige onderzoekspopulatie moeten worden ontworpen. Het doel van dit proefschrift was het verder ontwikkelen van (economische) evaluaties van geïntegreerde zorg. Hiervoor zijn de volgende onderzoeksdoelen bepaald:

1. *Het ontrafelen van de percepties van mensen met multimorbiditeit over wat een goede gezondheid en kwaliteit van zorg is*
2. *Het empirisch evalueren van een complexe integrale zorginterventie voor kwetsbare ouderen met behulp van Multi-Criteria Decision Analysis*

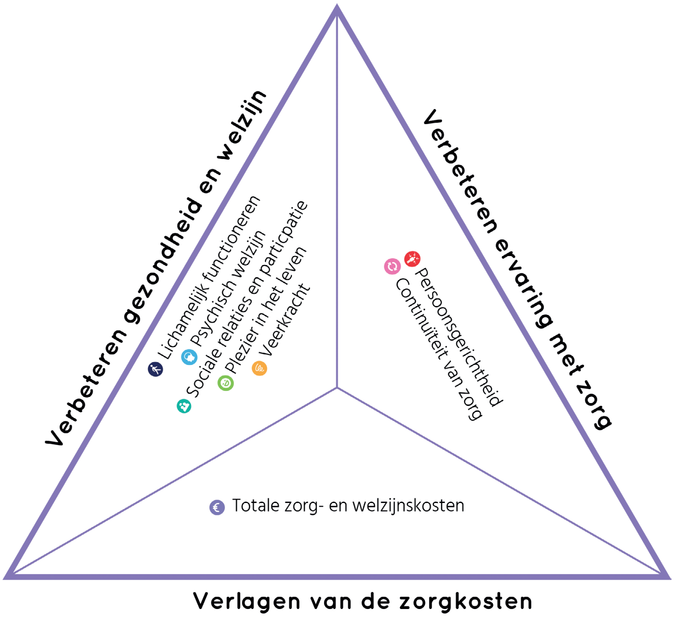
Deel 1. Het ontrafelen van de percepties van mensen met multimorbiditeit over wat een goede gezondheid en kwaliteit van zorg is

In het eerste deel van dit proefschrift is onderzocht wat goede gezondheid en zorg voor mensen met multimorbiditeit inhoudt, vanuit het perspectief van de patiënt zelf, en vanuit het perspectief van mantelzorgers, professionals, financiers en beleidsmakers.

Hoofdstuk 2 presenteert de resultaten van focusgroepen met personen met multimorbiditeit in acht Europese landen: Duitsland, Hongarije, Kroatië, Oostenrijk, Nederland, Noorwegen, Spanje en het Verenigd Koninkrijk. Deze kwalitatieve aanpak zorgde ervoor dat de perspectieven van personen met multimorbiditeit werden meegenomen in de discussie over zinvolle uitkomsten voor empirische evaluaties van integrale zorg. Deelnemers aan het onderzoek werd gevraagd om (1) te definiëren wat een goede gezondheid en welzijn, en een goed zorgproces voor hen betekende, en (2) om de belangrijkste uitkomstmaten te prioriteren. In alle landen legden de deelnemers veel nadruk op ‘plezier in het leven’, ‘vrijheid en onafhankelijkheid’, ‘psychisch welzijn’ en ‘het onderhouden van sociale relaties en contacten’. Bij het definiëren van een goed zorgproces hechtten de deelnemers de meeste waarde aan goed gecoördineerde en soepele overgangen tussen zorg (d.w.z. continuïteit van zorg). Dit kan worden verklaard door het feit dat patiënten met multimorbiditeit vaak te maken hebben met veel verschillende zorgverleners, organisaties en sectoren gedurende hun zorgtraject. Ook werden interacties met zorgverleners als belangrijk genoemd: een respectvolle behandeling, een holistische benadering, gedeelde besluitvorming en goede communicatie – zowel tussen zorgverlener en deelnemer als tussen zorgverleners onderling. Interessant om te noemen is dat aspecten gerelateerd aan de medische gezondheidstoestand en klinische indicatoren, zoals fysiek functioneren, in veel mindere mate werden genoemd. De studie concludeerde dat een brede reeks uitkomstmaten die betrekking hebben op mentale en fysieke gezondheid, welzijn, sociale relaties en procesindicatoren die de ervaring met zorgverlening weerspiegelen, moeten worden opgenomen in evaluaties van integrale zorgprogramma’s voor personen met multimorbiditeit.

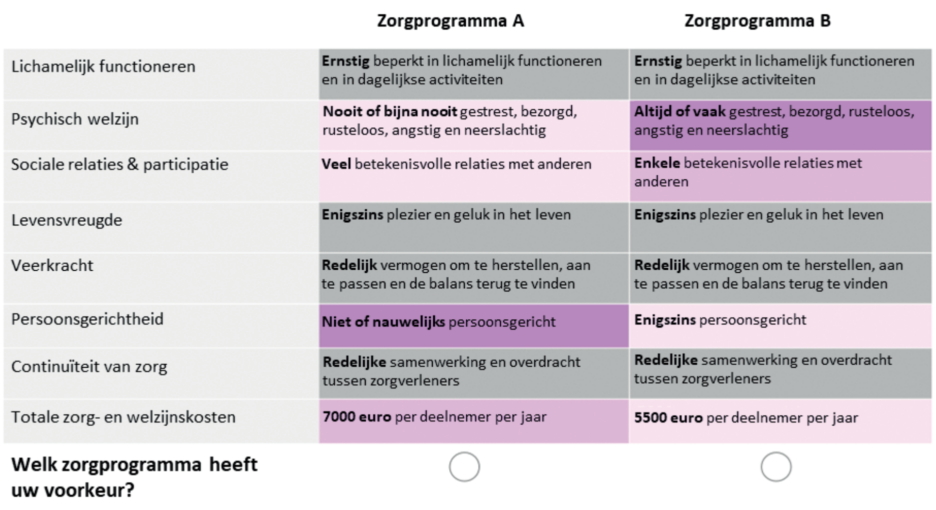
De resultaten uit hoofdstuk 2 speelden een belangrijke rol in de volgende hoofdstukken, aangezien deze resultaten de basis vormden voor de ‘*kernset* van uitkomstmaten’ die in het SELFIE-project werd opgesteld en die een centrale rol speelde in dit proefschrift. Hoewel de focusgroepen de basis hebben gelegd voor deze kernset, is er ook gebruik gemaakt van drie andere informatiebronnen: 1) een literatuuronderzoek uitgevoerd voor de ontwikkeling van een conceptueel raamwerk van integrale zorg, 2) bijeenkomsten met de nationale en internationale stakeholder adviesraden van het SELFIE-project met patiënten, mantelzorgers, professionals, financiers en beleidsmakers, en 3) een overzicht van de doelstellingen en uitkomstmaten die al werden gebruikt in de 17 SELFIE-casestudies (waaronder het Ketenzorgprogramma Kwetsbare Ouderen (KKO)). Deze vier bronnen resulteerden in de kernset van uitkomstmaten die de *Triple Aim* van integrale zorg omvat, zie **Figuur 1**.

Figuur 1. SELFIE Kernset van uitkomstmaten



Hoofdstuk 3 beschrijft de resultaten van een gepoolde analyse van de discrete-keuze-experimenten (DCE's) uitgevoerd onder patiënten, mantelzorgers, professionals, financiers en beleidsmakers. Het doel van dit hoofdstuk was tweeledig, namelijk, 1) onderzoeken in welke mate uitkomstmaten naast gezondheid worden gewaardeerd, en 2) de heterogeniteit bestuderen van voorkeuren voor uitkomstmaten van integrale zorg onder stakeholders die betrokken zijn bij integrale zorg. In de DCE kregen de respondenten keuzevragen voorgelegd waarin ze moesten kiezen tussen twee hypothetische integrale zorgprogramma's die verschilden in prestaties op de kernset van uitkomstmaten. Ze werden gevraagd om te kiezen welk zorgprogramma hun voorkeur heeft, waarbij ze afwegingen moesten maken tussen de verschillende uitkomstmaten van een integraal zorgprogramma. Het relatieve belang van elke uitkomstmaat was gebaseerd op de regressie-coëfficiënt van het beste niveau (niveau 3) gedeeld door de som van alle beste niveaus. Een voorbeeld van zo'n 'DCE-keuzetaak' is weergegeven in **Figuur 2**.

- **Figuur 2.** Voorbeeld van een DCE-keuzetaak

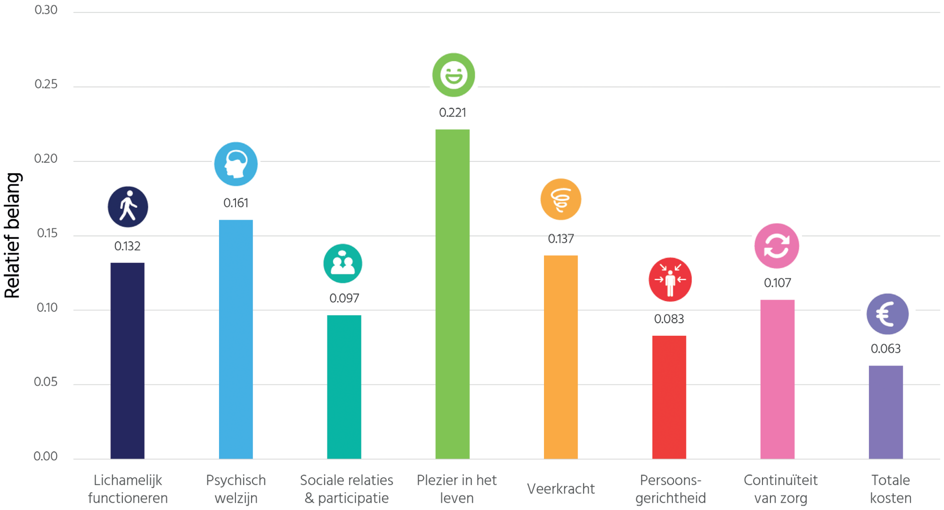


	Zorgprogramma A	Zorgprogramma B
Lichamelijk functioneren	**Ernstig** beperkt in lichamelijk functioneren en in dagelijkse activiteiten	**Ernstig** beperkt in lichamelijk functioneren en in dagelijkse activiteiten
Psychisch welzijn	**Nooit of bijna nooit** gestrest, bezorgd, rusteloos, angstig en neerslachtig	**Altijd of vaak** gestrest, bezorgd, rusteloos, angstig en neerslachtig
Sociale relaties & participatie	**Veel** betekenisvolle relaties met anderen	**Enkele** betekenisvolle relaties met anderen
Levensvreugde	**Enigszins** plezier en geluk in het leven	**Enigszins** plezier en geluk in het leven
Veerkracht	**Redelijk** vermogen om te herstellen, aan te passen en de balans terug te vinden	**Redelijk** vermogen om te herstellen, aan te passen en de balans terug te vinden
Persoonsgerichtheid	**Niet of nauwelijks** persoonsgericht	**Enigszins** persoonsgericht
Continuïteit van zorg	**Redelijke** samenwerking en overdracht tussen zorgverleners	**Redelijke** samenwerking en overdracht tussen zorgverleners
Totale zorg- en welzijnskosten	**7000 euro** per deelnemer per jaar	**5500 euro** per deelnemer per jaar
Welk zorgprogramma heeft uw voorkeur?	○	○

De gepoolde analyse van de DCE's toonde aan dat de top-3-uitkomsten met het hoogste relatieve gewicht bestond uit (1) plezier in het leven, (2) psychisch welzijn en (3) veerkracht. De uitkomstmaat 'totale zorg- en welzijnskosten' werd beschouwd als de minst belangrijke uitkomstmaat van een integraal zorgprogramma. **Figuur 3** geeft de gewichten van het relatieve belang weer voor de kernset van uitkomstmaten. De standaarddeviaties van alle uitkomstniveaus duidden op een grote variatie in voorkeuren onder de respondenten, wat de vraag opriep of er subgroepen met vergelijkbare voorkeuren te detecteren waren. Daarom werd latente klassenanalyse gebruikt om subgroepen van respondenten te identificeren (d.w.z. latente klassen) die vergelijkbare voorkeuren delen voor bepaalde uitkomstmaten. De latente klassen werden gevormd op basis van hun geobserveerde keuzegegevens. Vervolgens onderzochten we de kenmerken van de respondenten van elke subgroep.

De latente klassenanalyse identificeerde vier klassen die elk de nadruk leggen op specifieke uitkomsten: (1) kent een relatief hoger gewicht toe aan ervaring met zorg, (2) kent een relatief hoger gewicht toe aan plezier in het leven en psychisch welzijn, (3) kent een relatief hoger gewicht toe aan lichamelijke en geestelijke gezondheid, (4) ‹onverschillig›. Verschillen tussen de voorkeuren van de klassen was slechts zwak gerelateerd aan het stakeholderperspectief van respondenten – patiënt, mantelzorger, professional, financier of beleidsmaker – aangezien veel stakeholders aangaven dat ze meerdere rollen op zich nemen. Deze heterogeniteit in voorkeuren benadrukt het belang om bij het opzetten en evalueren van integrale zorgprogramma›s rekening te houden met een breed scala aan uitkomstmaten, waaronder welzijn en ervaring met zorguitkomsten.

Figuur 3. Relatieve belangrijkheidsgewichten voor de kernset van uitkomsten



Deel 2. Evaluatie van een complexe integrale zorginterventie voor kwetsbare ouderen

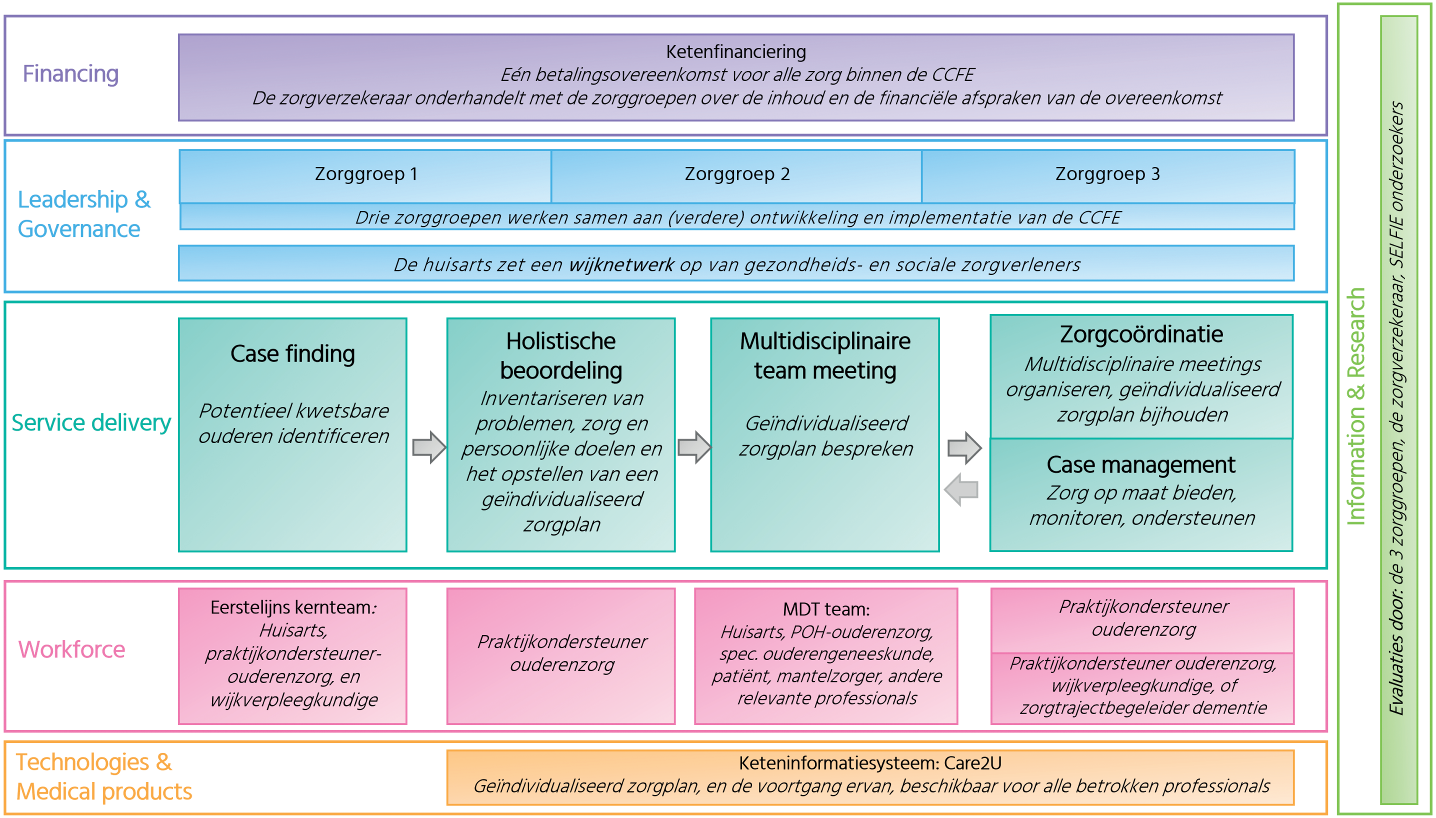
Het tweede deel van dit proefschrift richt zich op het empirisch evalueren van een integraal zorgprogramma, waarbij het Ketenzorgprogramma Kwetsbare Ouderen (KKO) als casestudy wordt genomen. De evaluatie omvatte het verkrijgen van een diepgaand begrip van het KKO en de beslissingscontext, het meten van de prestaties van het KKO en de controlegroep (gebruikelijke zorg) op de kernset van uitkomsten, en het combineren van deze prestaties met de relatieve belangrijkheidsgewichten, waarbij een MCDA werd uitgevoerd.

In **Hoofdstuk 4** wordt het KKO systematisch beschreven en geanalyseerd met behulp van een *thick description* (*NL = uitgebreide beschrijving*) methode die inzichten uit documentanalyse en kwalitatieve interviews combineert om een diepgaand inzicht van het programma te krijgen. Het doel was om de belemmerende en bevorderende factoren voor implementatie te identificeren en om innovatieve elementen van het KKO te onderscheiden, zodat deze inzichten het implementatieproces en het ontwerp van evaluatiestudies van soortgelijke programma's in de toekomst kunnen informeren. Er is gekozen voor *de thick description* methode omdat deze meerdere analyselagen omvat en verder reikt dan de beschrijvende aard van documentanalyse. Het maakt een grondig onderzoek van impliciete sociale relaties mogelijk, terwijl ook rekening wordt gehouden met de specifieke context van de interventie. De interviews met belangrijke stakeholders (bijvoorbeeld initiatiefnemers, patiënt, mantelzorger, huisarts, praktijkondersteuner) vulden de schriftelijke documenten aan door een dieper inzicht te geven in wat het programma onder de oppervlakte inhoudt en hoe het in de praktijk werkt. De resultaten zijn gestructureerd volgens het conceptuele raamwerk van integrale zorg ontwikkeld in het SELFIE-project, om integrale zorgprogramma's te beschrijven, ontwikkelen en implementeren. Het bestaat uit de volgende zes componenten die op macro-, meso- en microniveau aan bod kwamen, waarbij het individu centraal stond: 1) Service delivery, 2) Leadership & governance, 3) Workforce, 4) Financing, 5) Technologies & medical products, 6) Information & research.

Het KKO richt zich op thuiswonende kwetsbare ouderen met complexe zorgbehoeften en heeft tot doel persoonsgerichte zorgcoördinatie en casemanagement te bieden, zodat kwetsbare ouderen zo lang mogelijk thuis kunnen wonen. Het programma biedt meerdere belangrijke elementen die gebruikelijk zijn in programma's voor kwetsbare ouderen, bijvoorbeeld een holistische beoordeling van onvervulde gezondheids- en sociale zorgbehoeften, een individueel zorgplan, multidisciplinaire zorg, zorgcoördinatie en casemanagement. Het KKO onderscheidt zich echter van andere initiatieven door zich te richten op de kwetsbaarste huisartsenpopulatie, aanwezigheid van de patiënt en mantelzorger bij de multidisciplinaire zorgbijeenkomsten en de innovatieve financiering door middel van ketenfinanciering. Een samenvatting van het KKO en de meest onderscheidende elementen wordt gegeven in **Figuur 4**. We concludeerden dat de resultaten kunnen bijdragen aan de ontwikkeling van soortgelijke programma›s, aangezien de innovatieve componenten en succesfactoren van het KKO waarschijnlijk ook van toepassing zijn in een andere setting.

Hoofdstuk 5 presenteert de resultaten van de *Multi-Criteria Decision Analysis* van het KKO. Om de prestaties op de kernset van uitkomsten van het KKO en de gebruikelijke zorg te beoordelen, werden tijdens huisbezoeken gegevens verzameld onder 384 kwetsbare ouderen, op 0, 6 en 12 maanden door getrainde interviewers.

De resultaten na 6 maanden toonden aan dat het KKO beter presteerde dan de gebruikelijke zorg op het gebied van persoonsgerichtheid. Het fysiek functioneren nam echter af in beide groepen, maar nog meer in de interventiegroep. Ook na 12 maanden presteerde het KKO beter dan de gebruikelijke zorg op het gebied van persoonsgerichtheid, maar de verschillen tussen alle uitkomsten waren klein, zie **Figuur 5**. Vervolgens werden de prestatiescores vermenigvuldigd met de relatieve belangrijkheidsgewichten vanuit de vijf stakeholderperspectieven. De MCDA-resultaten lieten zien dat na 6 maanden, het KKO werd verkozen boven de gebruikelijke manier van zorg verlenen aan kwetsbare ouderen. Na 12 maanden lieten de MCDA-resultaten weinig verschillen zien tussen het KKO en de gebruikelijke zorg vanuit het perspectief van patiënten, mantelzorgers en professionals, terwijl financiers en beleidsmakers de



• **Figuur 4.** Resultaten *thick description* van het KKO volgens de zes componenten van het SELFIE Raamwerk

Financing

Ketenfinanciering
Eén betalingsovereenkomst voor alle zorg binnen de CCFE
De zorgverzekeraar onderhandelt met de zorggroepen over de inhoud en de financiële afspraken van de overeenkomst

Leadership & Governance

Zorggroep 1 | Zorggroep 2 | Zorggroep 3

Drie zorggroepen werken samen aan (verdere) ontwikkeling en implementatie van de CCFE

De huisarts zet een ***wijknetwerk*** *op van gezondheids- en sociale zorgverleners*

Service delivery

Case finding
Potentieel kwetsbare ouderen identificeren

Holistische beoordeling
Inventariseren van problemen, zorg en persoonlijke doelen en het opstellen van een geïndividualiseerd zorgplan

Multidisciplinaire team meeting
Geïndividualiseerd zorgplan bespreken

Zorgcoördinatie
Multidisciplinaire meetings organiseren, geïndividualiseerd zorgplan bijhouden

Case management
Zorg op maat bieden, monitoren, ondersteunen

Workforce

Eerstelijns kernteam:
Huisarts, praktijkondersteuner-ouderenzorg, en wijkverpleegkundige

Praktijkondersteuner ouderenzorg

MDT team:
Huisarts, POH-ouderenzorg, spec. ouderengeneeskunde, patiënt, mantelzorger, andere relevante professionals

Praktijkondersteuner ouderenzorg

Praktijkondersteuner ouderenzorg, wijkverpleegkundige, of zorgtrajectbegeleider dementie

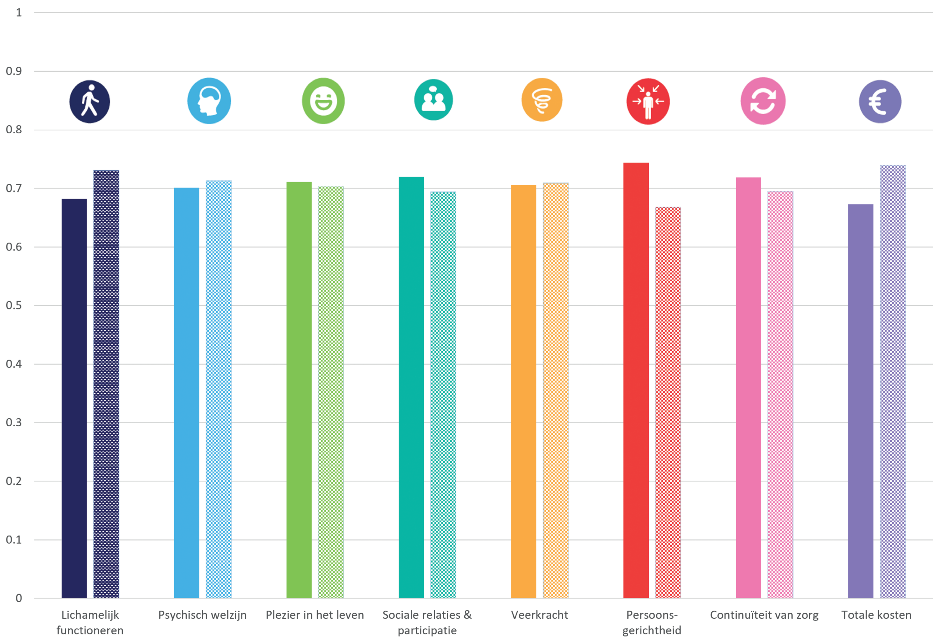
Technologies & Medical products

Keteninformatiesysteem: Care2U
Geïndividualiseerd zorgplan, en de voortgang ervan, beschikbaar voor alle betrokken professionals

Information & Research

Evaluaties door: de 3 zorggroepen, de zorgverzekeraar, SELFIE onderzoekers voorkeur leken te geven aan gebruikelijke zorg. Concluderend, het KKO kan de voorkeur hebben als plezier in het leven en persoonsgerichtheid belangrijker worden geacht dan kosten en fysiek functioneren.

- **Figuur 5.** Gestandaardiseerde prestatiescores van het KKO en gebruikelijke zorg na 12 maanden



Voetnoot: Links = het KKO, rechts = gebruikelijke zorg. Resultaten zijn gestandaardiseerd, wat betekent dat ze allemaal op de schaal van 0-1 liggen en een hogere score duidt op een betere prestatie (voor kosten betekent een betere prestatie lagere kosten).

Verder pleiten we voor een breder gebruik van MCDA voor het evalueren van veelzijdige integrale zorg met een verscheidenheid aan beoogde resultaten die verder reiken dan kwaliteit van leven en levensverlenging. Deze aanpak kan dan nuttige informatie opleveren om een transparant, consistent, verantwoordelijk, geloofwaardig en acceptabel besluitvormingsproces van op waarde gebaseerde zorg mogelijk te maken.

Hoofdstuk 6 duikt dieper in de toepasbaarheid van MCDA door te onderzoeken hoe MCDA de kosteneffectiviteitsanalyse (KEA) kan aanvullen om besluitvorming op lokaal niveau te ondersteunen. Daarnaast introduceert dit hoofdstuk de *Conditional Multi-attribute Acceptability Curve* (CMAC), die de kans weergeeft dat het KKO de hoogste *overall value score* heeft (d.w.z. de voorkeur heeft boven gebruikelijke zorg), terwijl het budget (gegeven een bepaalde doelgroepgrootte) onder een vastgestelde drempel voor het beschikbare budget blijft. Uit de resultaten van de KEA, met een drempelwaarde voor de betalingsbereidheid van 50.000 euro per *QALY (Quality-Adjusted Life Year; NL = voor kwaliteit van leven gecorrigeerd levensjaar)*, bleek dat het KKO waarschijnlijk niet kosteneffectief is. Vanuit een MCDA-perspectief heeft het KKO echter aangetoond dat het potentieel even effectief en betaalbaar is als de gebruikelijke zorg, terwijl het ook het plezier in het leven en persoonsgerichtheid verbetert. Daarom kan de investeringsbeslissing op basis van de twee benaderingen verschillen wanneer beleidsmakers ervoor kiezen om innovaties te financieren die meer waarde bieden, zoals gemeten met een brede reeks uitkomstmaten, waaronder plezier in het leven en persoonsgerichtheid. MCDA is daarmee een aanvulling op de KEA doordat het besluitvormers een uitgebreider beeld geeft van de effectiviteit van een integraal zorgprogramma, bijvoorbeeld in het kader van de Triple Aim van integrale zorg. Het hangt af van de beslissingscontext of KEA of MCDA moet worden toegepast om beslissingen te informeren. Conventionele KEA is wellicht het meest geschikt voor besluitvorming op nationaal niveau met betrekking tot de keuze welke behandelingen aan het basispakket zorg moeten worden toegevoegd, aangezien het een directere vergelijking tussen ziektegebieden en behandelingen mogelijk maakt. Het is echter mogelijk dat een KEA niet volledig de impact weergeeft van integrale zorg voor kwetsbare ouderen, omdat deze interventies vaak gezondheids- en sociale zorg combineren en gericht zijn op het verbeteren van uitkomsten die verder gaan dan gezondheid. MCDA kan dan informatie over de waarde van integrale zorginterventies aanvullen, wat vooral nuttig kan zijn bij besluitvorming op lokaal niveau. Deze bevindingen zijn relevant, omdat het de acceptatie en implementeerbaarheid van innovaties in de ouderenzorg kan bevorderen.

Ten slotte werden de belangrijkste bevindingen van dit proefschrift samengevat en besproken. Het laatste hoofdstuk reflecteert op de theoretische en methodologische overwegingen en werpt licht op de implicaties voor beleid, praktijk en onderzoek.


8

List of publications and conferences

LIST OF PUBLICATIONS

Czypionka, T., Kraus, M., Reiss, M., Baltaxe, E., Roca, J., Ruths, S., Stokes, J., Struckmann, V., Tandara Haček, R., Zemplenyi, A., **Hoedemakers, M.**, & Rutten-van Mölken, M. (2020). The patient at the centre: Evidence from 17 European integrated care programmes for persons with complex needs. *BMC Health Services Research, 20*. https://doi.org/10.1186/s12913-020-05917-9

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https://doi.org/10.1055/a-1547-6898

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SELFIE Steering Committee (SSC) Meetings and presentations

SSC Meeting Manchester (United Kingdom) **2016**

Poster presentation:

“Work Package 2 poster: The Netherlands: Care Chain Frail Elderly”

SSC Meeting Barcelona (Spain) **2017**

Presentation:

“Work Package 8: Dissemination and communication of results”

Poster presentation:

“Work Package 4 & 5 poster: The Netherlands: Study design Care Chain Frail Elderly”

SSC Meeting Bergen (Norway) **2017**

Presentation:

“Work Package 4: Weight elicitation study: progress recruitment professionals, payers and policy makers”

Poster presentation:

“Work Package 5: Progress report Netherlands: Care Chain Frail Elderly”

SSC Meeting Budapest (Hungary) **2017**

Presentation:

“Work Package 5: Progress cost calculations of the integrated care programs: CCFE”

Poster presentation:

“Work Package 5: Progress and results evaluation studies WP5: CCFE”

SSC Meeting Berlin (Germany) **2018**

Presentation:

“Work Package 8: SELFIE Final Conference Brainstorm”

(Inter) National Conferences

JA-CHRODIS Nivel - Invitational conference Utrecht (NL) **2017**

- Betere zorg voor mensen met multimorbiditeit
  ENG: Better care for people with multi-morbidity

NCVGZ (Nederlands Congres Volksgezondheid) Amersfoort (NL) **2017**

- De professional maakt het verschil
  ENG: The professional makes the difference

Oral presentation:
“Who does what? 'New' professional roles in integrated care programs for people with multimorbidity”
Poster presentation:
“The SELFIE Project: Integrated care for persons with multi-morbidity”

International Conference on Integrated Care (ICIC) Dublin (Ireland) **2017**

- Building a platform for integrated care: delivering change that matters to people

Oral presentation:
“How to deliver care to frail elderly?
A SELFIE research on the barriers and facilitators in the Netherlands”

NCVGZ (Nederlands Congres Volksgezondheid) Utrecht (NL) **2018**

- Preventie als mindset
  ENG: Prevention as mindset

International Conference on Integrated Care (ICIC) Utrecht (NL) **2018**

- Value for People and Populations: Investing in Integrated Care

Oral presentation:
“Evaluating an integrated care programmefor frail elderly using Multi-Criteria Decision Analysis The design of a case study in SELFIE”
Poster presentation:
“Strengthening the evidence-base of integrated care in multi-morbidity: 17 Multi-Criteria Decision Analyses (MCDA) in eight European countries”

European Health Economics Association (EuHEA) Maastricht (NL) **2018**

- Shaping the Future: the Role of Health Economics

Oral presentation:
“Importance of Triple Aim outcome measures: do patients, partners, professionals, payers and policy makers differ in opinion?”
in Workshop: “Strengthening the evidence-base of integrated care for people with multi-morbidity in Europe using Multi-Criteria Decision Analysis (MCDA) –The SELFIE project”

SUSTAIN Final Conference Brussel (Belgium) **2019**
- Sustainable Tailored Integrated Care for Older People in Europe

International Conference on Integrated Care (ICIC) San Sebastian (Spain) **2019**
- Evaluating and implementing models of integrated people-centred services

Oral presentation:
"Evaluating an integrated care programme for frail elderly using Multi-Criteria Decision Analysis - The results of a case study in SELFIE"

International Conference on Integrated Care (ICIC) Virtual conference **2020**
- Shared Vision and Values for Integrated Care

Low Lands Health Economic Study Group (LoLa HESG) Maastricht (NL) **2022**
Presentation:
"Moving beyond QALYs in elderly care: how can Multi-Criteria Decision Analysis complement Cost-Effectiveness Analysis in local level decision-making"


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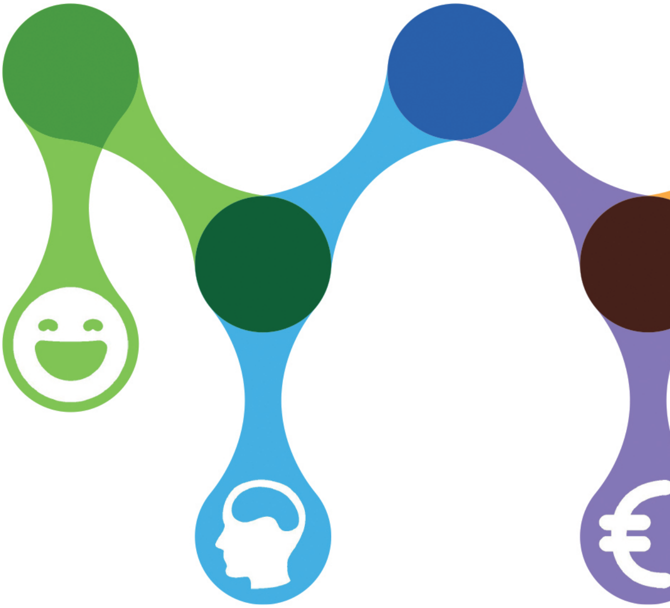
About the author

ABOUT THE AUTHOR

Maaike Hoedemakers was born in Rotterdam, the Netherlands on January 2, 1990. In 2013, she obtained a bachelor's in Communications at the Rotterdam University of Applied Sciences. Alongside her studies, she worked at *Laurens*, specialised in elderly care, rehabilitation, and nursing home care, (from 2005-2014) which sparked her interest in healthcare. In 2015 she obtained her master's degree in Health Economics, Policy and Law, from the Erasmus University Rotterdam. During her master's, she specialised in Health Technology Assessment. After graduation, she started as a PhD candidate at the Erasmus School of Health Policy & Management under supervision of prof. dr. M. Rutten-van Mölken and dr. A. Tsiachristas, which resulted in this thesis. Her PhD project was part of the European Union's Horizon 2020 project SELFIE (**S**ustainable Int**e**grated Care Mode**l**s for Multi-Morbidity: Delivery, **F**inanc**i**ng and P**e**rformance) and mainly focused on improving the evaluation of integrated care for persons with multimorbidity using Multi-Criteria Decision Analysis (MCDA).



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