Real-world Implementation of Value-based Healthcare

Evaluation from a patient, healthcare organization and health policy perspective



Fiona Koster

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Printed by: Print Service Ede – The Netherlands

ISBN: 978-90-834766-4-3

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Real-world Implementation of Value-based Healthcare

Evaluation from a patient, healthcare organization and health policy perspective

Implementatie van waardegedreven zorg in de praktijk: evaluatie vanuit een patiënt, gezondheidszorgorganisatie en beleidsperspectief

> Thesis to obtain the degree of Doctor from the Erasmus University Rotterdam by command of the rector magnificus

> > Prof.dr.ir. A.J. Schuit

and in accordance with the decision of the Doctorate Board. The public defence shall be held on

> Wednesday 27 November 2024 at 13.00 hrs by

Fiona Koster

born in Den Helder.

Frafing

Erasmus University Rotterdam

Doctoral Committee

| Promotor: | Prof.dr. A.E.A.M. Weel-Koenders |
|----------------------|--|
| Other members: | Prof.dr.ir. C.T.B. Ahaus Prof. P.B. van der Nat |
| | Prof. dr. C.A. Uyl-de Groot |
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"Je gaat het pas zien als je het doorhebt"

- Johan Cruijff -



Chapter 1

General Introduction

1

At a certain point in life, every person will be exposed to healthcare, either as a patient or (increasingly) as a caregiver. Due to the fact that healthcare systems are complex, involving a variety of stakeholders and organizations, patients can easily feel overwhelmed when entering this system [1]. As a patient, the main goal of visiting healthcare providers and receiving healthcare services, is obtaining a desired state of health. Healthcare systems that want to be effective should prioritize the needs of the patient [2]. Hence, the delivery of care should ideally be organized around the patient, taking into account the preferences and values as obtained through healthcare services [2]. However, in practice, care is traditionally organized from the perspective of the provider or the policy maker [2]. As a result, the current system of care delivery often revolves around the healthcare providers [2]. Furthermore, providers are usually reimbursed by the number of performed services and therefore providers have a financial incentive to supply health services, irrespective of whether they are essential, preferred or congenial [3]. This fee-for-service (FFS) reimbursement system can undermine clinical accountability, since providers get paid regardless of the need for (or quality of) their services, driving up healthcare costs [4]. Moreover, this provider oriented (reimbursement) structure interferes with the issues encountered from a macroeconomic perspective, as the necessity to contain the growth of healthcare expenditures is of importance to decision makers. Healthcare costs in the Netherlands have been increasing at a disproportionate rate compared to GDP growth for several decades: they increased from 7.7 per cent of GDP in 2000 to 11.2 per cent in 2021 [5]. The Dutch care sector employs around 1.4 million people or some one sixth of the work force. The need to address this issue has led to a heightened policy interest in transforming the healthcare delivery system [6]. To align the organization of healthcare (delivery) with the patients' preferences, taking into account the perspective of the healthcare organizations (i.e. providers) and policy makers, the concept of value-based healthcare (VBHC) can be used as a stepping stone to accomplish this goal.

Value-based healthcare (VBHC)

VBHC was introduced in 2006 by Michael Porter and Elizabeth Teisberg [7]. VBHC aims to sustainably improve the outcomes that matter to people by organizing care around patients with a specific condition, standardizing outcome and cost measurement, and then using this data to monitor and compare performance within and between organizations, with the aim of stimulating learning and improvement [7]. In this system, providers are incentivized to help patients improve their health, prevent chronic diseases, and adopt evidence-based practices to prioritize patient health improvements, disease prevention, and evidence-based practices over the volume of services provided [7]. This approach contrasts with the traditional fee-for-service or capitated payment models, which reimburse providers based on the volume of the services they provide [7]. The "value" in value-based healthcare comes from measuring the health outcomes achieved weighed against the costs of the provided services [7]. The VBHC approach has gained popularity worldwide [6]. The Dutch healthcare system has placed a greater emphasis on VBHC, following the global trend towards VBHC adoption. Formal institutions such as the Dutch Healthcare Institute, have included VBHC as a crucial component in their vision and founded the Linnean Institute to promote that approach [8]. The government is also engaged in a variety of programs to facilitate the implementation of outcomeoriented care, which emphasizes the concept that choosing the optimal treatment for a disease should also be one that suits the individual patient [9]. To provide patients with the most optimal treatment, shared-decision making is promoted by the Dutch government. As a consequence, healthcare delivery becomes more patient-oriented, allowing the patient to have a more significant involvement in care delivery [10]. In the end, the transformation to a value-orientated system contributes to a more resilient and efficient healthcare delivery system.

Aim

The aim of this thesis is to research options to integrate the patient perspective with both the healthcare organization and policy maker perspective within the VBHC concept. This thesis serves as a link between the theoretical constructs of VBHC and its empirical implementation within the healthcare framework of the Netherlands, delineating the capacity of the VBHC model to improve the management of (chronic) conditions such as Rheumatoid Arthritis (RA). The focus of this thesis is specifically on the cost side of the VBHC equation, by applying a health economic view on the steps of the value agenda. A real-world setting is examined to provide insight in the practical implementation of VBHC. Three perspectives are considered in this thesis; the micro, meso and macro level. The micro level concerns the patient perspective, the meso level represents the healthcare organization and the macro level the health policy perspective.

Rheumatoid arthritis

In this thesis, Rheumatoid Arthritis (RA) is the medical condition of interest. RA is the most prevalent form of the rheumatoid diseases. The specific cause of RA is still unknown; however, it has become evident that a combination of genetics and environmental components is responsible for developing the disease [11]. RA is a chronic inflammatory disease predominantly affecting the joints of patients [11]. As a result of the inflammations, their physical functioning declines, also impacting the quality of life of patients in terms of productivity and participation in society [11]. Approximately 1.5% of the Dutch population suffers from RA and due to the ageing of the population

this percentage is expected to increase [11]. Disease activity is often the target in the treatment of RA patients. Disease activity can for example be measured as a DAS28CRP score; a measure assessing the 28-joint count and the C-reactive protein (CRP) levels in the blood, with scores ranging from 0 to 9.4 [12]. According to the DAS28CRP score, a value of 2.6 or higher indicates a patient is in a state of inflammation and a score below 2.6 indicates a patient is in remission, i.e. experiencing no significant joint inflammation [12]. First-line treatment in achieving remission or a low(er) disease activity is via the administration of Disease Modifying Anti-Rheumatic Drugs (DMARDS) [12]. Early diagnosis is crucial, as starting the treatment promptly is beneficial in reaching and preserving a state of remission [12]. Over the past decades, the management of RA has improved, with the approval of biological disease-modifying anti-rheumatic drugs (bDMARDs) targeting the tumor necrosis factor (TNF). Although treatment has evolved and the burden on patients decreased, RA remains a chronic illness and therefore patients require lifelong treatment including outpatient visits and medication. However, to reduce the challenges patients face in accessing healthcare services, e-health has experienced a surge in popularity [13]. E-health can provide a solution for chronic patients as self-management is enhanced and the number of physical consultations can be reduced [13]. Hence, embracing e-health also facilitates the transition to providing care at the most appropriate place as preferred by the patient.

Value-based healthcare implementation

To implement VBHC in practice, Porter and Teisberg developed a value agenda, a model to implement VBHC (figure 1) [14]. The model consists of six components, from organizing healthcare in an integrated practice unit to building an integrated information technology platform [14]. All six components can contribute to the restructuring of the healthcare system.

Figure 1. The value agenda [7]



1. Organize into Integrated Practice Units

As recommended by the value agenda, this thesis will start with describing the actual care delivery process as a first step to organize healthcare into an Integrated Practice Unit (IPU). The Care Delivery Value Chain (CDVC), a framework assisting in the understanding of the arrangement and structure of medical care services for specific conditions, was applied to provide insight in the delivery process.

2.1 Measure outcomes for every patient

In clinical practice, the first action in implementing VBHC is often the measurement of outcomes. Porter suggested categorizing outcomes at three levels, labeled as tiers [15]. The first tier evaluates the health status achieved (or retained) [15]. The second tier considers the process of recovery and the third, the sustainability of the patient's health condition [15]. From a VBHC perspective, outcomes that matter to patients are the only authentic measures of quality. That perspective requires the assessment of Patient Reported Outcome Measures (PROMs). PROMs are questionnaires that provide insight in the wellbeing of a patient, not limited to clinical parameters, but encompassing the overall functioning of a patient [16]. Examples of domains incorporated in these questionnaires, are fatigue and physical functioning. To promote standardized and comprehensive outcome measurement at a global level, 2012 saw the foundation of the International Consortium for Health Outcomes Measurement (ICHOM) [14]. ICHOM utilizes international data and best practices of healthcare providers to develop minimum outcome sets for several medical conditions [14]. Currently, 40 outcome sets are available via ICHOM, that include standardized outcome questionnaires, assessment

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instruments and frequency of administrating a questionnaire. This thesis uses PROMs as defined by ICHOM, to measure health outcomes. PROMs are either generic or disease specific [16]. Generic PROMs are intended to evaluate a patient's overall health not related to any particular illness, such as Euro Quality of life - 5 Dimension (EQ-5D), 36-Item Short Form Health Survey (SF-36) and the Patient-Reported Outcomes Measurement Information System (PROMIS). On the other hand, disease specific PROMs are tailored to evaluate health aspects that are specific to a particular disease [16]. Despite the fact that PROMs are increasingly applied in clinical practice to evaluate the healthcare results obtained, the initial purpose was to employ PROMs in research [16]. As part of this thesis, PROMs will be assessed to evaluate a patient-reported and clinical outcome dashboard that is carried out in the clinical practice.

2.2 Measure cost for every patient

The value equation integrates both components of the second step of the value agenda, which involves dividing the outcomes achieved, as measured by the change in a PROM, by the total cost of the associated care [7]. Although numerous studies have researched the nominator side of the value equation, i.e., the (patient-reported) outcomes and experiences, there is limited knowledge regarding the cost aspect. However, to transform the healthcare system from a volume to a value-based system, insight in the cost side of the value equation is necessary. In healthcare, a distinction can be made between the costs of care and the reimbursements received by the care providers from health insurers. The emphasis in VBHC should be on the former since reimbursements do not accurately reflect the actual costs as incurred by a healthcare organization. This thesis wants to contribute to the practice of VBHC, by providing evidence on the implications of including the cost aspect of the value equation.

Costing accounting in healthcare

As part of the second component of the value agenda, measuring outcomes and costs for every patient, insight in the costs of care is relevant [14]. To estimate the full costs of a care cycle, cost accounting methods can provide a solution. Cost accounting is the process of calculating the costs to provide a service or to manufacture a product [17]. To illustrate cost accounting, the example of a total hip replacement can be used. A total hip replacement consists of several components, e.g., the diagnostic part of the treatment and the surgery. The initial step is to determine a cost object to assign costs to [17]. For these cost objects, direct and indirect costs can be distinguished [17]. Direct costs are expenses that can be directly linked to the cost object [17]. In case of a total hip replacement, the costs of the surgeon and the hip prosthesis are directly linked to the surgery. With respect to indirect costs, cost allocation is more complicated [17]. In the example of the hip replacement, the operating room can be considered as an indirect cost: the use of the operating room is not limited to hip replacement surgeries but can also be used for other medical procedures.

Methodologies

Cost accounting within healthcare has developed from traditional costing to activitybased costing (ABC) and, since 2003, to time-driven activity-based costing (TDABC) (figure 2) [18]. In traditional costing, indirect costs are allocated to cost objects based on generic keys, such as a surcharge on direct expenses, cost reimbursements or staff numbers [18]. ABC ventures to link indirect costs to factors, such as support services, that drive those expenses. Hence, a medical procedure (cost object) that requires a large amount of time in for example the operating room, is allocated more of the associated expenses [18]. The transition to ABC increased the accuracy of the indirect cost allocation, since indirect costs were allocated based on the volume of services that drive those expenses [18]. However, a major drawback of ABC is the cost of creating and maintaining a model for a complex organization, such as a hospital [18]. Time-driven activity-based costing (TDABC) offers a simplification of the ABC-method, by directly estimating the resource demands of each cost object, rather than assign costs first to activities and then to cost objects [18]. Within healthcare that approach allows to integrate the patient perspective in the calculations, because the costs associated with each stage of the patient's journey can be computed.

Implementing TDABC with patient-centered insights

For example, applying TDABC to the total hip replacement would involve quantifying the time spent on each component of the procedure - from diagnostics to surgery - and assigning costs accordingly. This method not only offers insights into the cost structure of a patient's care cycle but also ensures that financial considerations remain closely tied to patient outcomes.

Figure 2. Development of cost accounting



3. Move to bundled payments for conditions

For VBHC to be implemented effectively among integrated and multidisciplinary teams, there is a need for supportive payment models [6]. Bundled Payments (BPs) are proposed as an alternative payment model, that comprises of a fixed payment that is predetermined for a group of providers, covering all care services associated with a specific treatment or medical condition within a designated time frame [19]. BPs can support the transition to a VBHC system since care providers are rewarded on basis of outcomes achieved across the episode of care rather than the number of care services provided [19]. Therefore, BPs transfer (part of) the financial risk related to healthcare provision to healthcare providers, holding them jointly responsible for both the cost and quality of care [20]. This approach also provides them with incentives and the flexibility to work together and coordinate care across various healthcare platforms [20]. A common challenge for BPs is setting the price for a bundle of services. Due to the variation in health risks in the patient populations served, resulting in differences in expected healthcare expenditures, there is a need to account for variations in healthcare conditions [20]. Thus, to drive a common understanding concerning the approaches used in BPs to account for the difference in health risks of patients, a systematic review forms part of this thesis.

4. Integrate multi-site care delivery systems

By conducting economic evaluations, the process as described in step four of the value agenda, integrate care delivery systems, can be provoked [14]. The integration of multisite care delivery systems, builds on the knowledge of integrating systems to eliminate fragmented care and optimize the types of care provided at each location, creating significant opportunities for enhancing value in healthcare delivery. To assess the value enhancement of an integrated intervention compared with the traditional care provision, an economic evaluation based on PROMs is explored in this thesis. Economic evaluations are predominantly conducted to determine the value on a macro policy level. However, with the common factor of patient value as defined by the PROMs, the micro and meso perspective can be incorporated in the economic evaluation.

Economic evaluations in healthcare

In an economic evaluation, health outcomes and costs can be combined to analyze the value of alternative courses of action. Traditionally, economic evaluations are performed to determine the value of interventions and technologies such as a new medicine, vaccine or MRI scanner. From a VBHC perspective, economic evaluations can be used to deliver (better) patient-reported outcomes at a lower total cost. VBHC adds on the knowledge of economic evaluation due to the broader perspective of the evaluations. VBHC allows to evaluate the standard of care and innovations. By combining VBHC and the traditional economic evaluation, procedures can be identified that may not offer enough benefit to justify their cost and could therefore be discontinued.

Four types of economic evaluations can be distinguished in healthcare, the cost minimization analysis, cost benefit analysis, cost-effectiveness analysis and the costutility analysis. A cost minimization analysis compares the costs of alternative methods, but the health outcomes are not taken into account. In a cost benefit analysis, both costs and the value of health outcomes are measured in monetary terms. The cost-effectiveness and cost-utility analysis are quite similar, however effects in the cost-effectiveness analysis are measured in natural units (e.g. number of CVAs prevented), whereas a cost-utility analysis measures outcomes in Quality-Adjusted Life Years (QALYs).

5. Expand geographic reach

The value agenda states that in order to accomplish the integration of VBHC and increase value on a broad scale, exceptional providers of care for specific medical conditions must serve a significantly larger number of patients by strategically expanding their outstanding IPUs [14]. Geographical expansion should concentrate on enhancing value, rather than merely boosting patient numbers [14]. Because of the increasing number of people with two or more chronic conditions, concentration in terms of volume of care while improving value is complex. To shed light on the prevalence of multi-morbidity and related healthcare utilization, the difference when considering a single-hospital perspective and population perspective is examined in this thesis.

6. Build an Enabling Information Technology Platform

The first five components of the value agenda can benefit from a sixth component, which is an effective information technology (IT) platform [14]. IT systems can encourage the different parts of an IPU to collaborate, facilitate measurement and innovative reimbursement strategies and unite the various components of a well-designed delivery system [14]. To enable proper measurement of outcomes, a well-designed IT platform is fundamental.

Additionally, adequate registration of the delivered healthcare, enhancing transparency of the reimbursement and financing of healthcare expenditures is important. Thus, when viewed through a VBHC lens, making an accurate diagnosis helps to ensure that patients receive timely and tailored treatment that aligns with their needs. This, in turn, supports the overarching objective of enhancing patient-centered care and improving health outcomes.

Data sources

The analyses performed in this thesis, except for the systematic review in chapter 8, are all based on real-world data. The majority of the data are obtained from the rheumatology and clinical immunology department at Maasstad Hospital, a topclinical teaching and research hospital in Rotterdam, the Netherlands. Over 3,000 RA patients are treated annually at the outpatient department. Data are processed anonymously in the conducted analyses. By means of the JOINT Evaluation study, researchers of the rheumatology and clinical immunology department are authorized to evaluate (financial) data of RA patients retrospectively. The data encompasses demographics such as age and gender. Furthermore, clinical outcomes can be examined, e.g., diagnoses, laboratory results and radiographic imaging can be retrieved. And in financial data, cost prices, index year and performed care activities can be assessed.

Real-world RA cohort

The JOINT Compassion study protocol comprises the scientific analyses with respect to the PROMs that are routinely collected as part of our standard rheumatology care since 2017. The examined PROMs are derived from the ICHOM standard set for Inflammatory Arthritis (IA) [21] and filled out approximately every 6 months. The five questionnaires included in the standard set are Functional Assessment of Chronic Illness Therapy (FACIT), Health Assessment Questionnaire-Disability Index (HAQ-DI), Rheumatoid Arthritis Impact of Disease (RAID), Euro Quality of life -5 Dimension (EQ - 5D), the Work Productivity and Activity Impairment (WPAI) and the Dutch Quality registry Rheumatoid Arthritis (DQRA) questionnaires. As of May 2023, this cohort consisted of 1377 RA patients from whom multiple/repeated PROMs and PREMs data are available.

Statistics Netherlands

In addition to the datasets from Maasstad Hospital, Statistics Netherlands data were used to compare healthcare costs of the Dutch population with the single-hospital population of Maasstad Hospital (Chapter 5).

Outline

The steps of the value agenda can be defined at the micro, meso and macro structure.

Part 1: Patient level (micro)

Within VBHC, healthcare delivery should enhance the value as perceived by the patient. Thus, the first part of this thesis focusses on evaluating VBHC on a patient level. The starting point, in line with the first step of the VBHC implementation model, is to provide insight in the medical condition around the patient. Hence, in chapter 2, the Care Delivery Value Chain and the related patient journey for RA patients is mapped. In chapter 3, the value chain framework presented in chapter 2 is used as a basis to calculate the total costs of the full cycle of care. To calculate the RA cycle costs, time-driven activity-based costing is combined with the theory of fuzzy logic.

Part 2: Healthcare organization level (meso)

The healthcare organization perspective (meso level) will be evaluated in chapters 4, 5 and 6. In chapter 4, the (financial) impact of misclassification of the RA diagnosis will be researched, since accurate data at the patient level is necessary to evaluate the incurred costs and effects. As patients with chronic diseases such as RA have an increased probability of suffering from other diseases, the chance of enhanced expenditures within multi-morbid patients is also higher. To analyze the impact of multi-morbidity on healthcare organizations, a Dutch population perspective is compared with the single-hospital perspective in chapter 5. Chapter 6 demonstrates the effects of patientreported outcomes on healthcare utilization, by assessing a patient-reported outcomes dashboard in the hospital.

Part 3: Health policy level (macro)

The macro perspective is described in chapters 7 and 8, in which healthcare decision making and an alternative payment model are discussed. To incorporate the patient

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perspective in an economic evaluation, PROMs are applied to evaluate the costeffectiveness of electronic consultations in chapter 7. Chapter 8 gives an overview of the approaches available to account for patient risk profiles in an alternative payment model (i.e., bundled payments), designed to move from a volume-based to value-based reimbursement model.

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Part 1

Micro level: patient



Chapter 2

Defining the Care Delivery Value Chain and Mapping the Patient Journey in Rheumatoid Arthritis

Koster F, Lopes Barreto D, Nair SC, Kok MR and Weel-Koenders AEAM

Rheumatology International 2022 Sep 23:1–8. doi: 10.1007/s00296-022-05215-z

Abstract

Rheumatoid Arthritis (RA) is a chronic disease that impacts patients' quality of life. Sophisticated organization of care delivery drives quality improvement. Therefore, the study objective was establishing a validated process map of the care cycle for RA patients. Hence, increasing transparency and optimizing care delivery and identifying areas of improvement. To map the RA care cycle, the care delivery value chain (CDVC) approach was used as framework to document activities and resources systematically. A mixed method study was conducted where quantitative data on activities were collected from health records and unstructured interviews with medical staff were held. Consequently, the process map was separately validated in a consensus meeting with a delegation of the medical staff and patient advisory board. At the start of the care cycle, the focus is predominantly on defining the treat-to-target strategy and examining disease activity. Towards the monitoring phase, tapering medication and managing the disease through patient-reported outcome measures are becoming increasingly important. Although patient's functioning, quality of care and patient's evaluation of received care are monitored, reflection of CDVC and engaging patients in the evaluation process resulted in improvement actions on outcome and process level. Mapping the RA care cycle following a systematic approach, provides insight and transparency in delivered activities, involved resources and the engagement of patients and caregivers at multiple levels, contributing to a system facilitating value-based care delivery. The CDVC framework and applied methodology is recommended in other conditions. Future research will focus at assigning outcomes and costs to activities and evaluating interventions to explore patient value.

2

Introduction

Rheumatoid Arthritis (RA), the most common form of rheumatic disease, is an autoimmune illness affecting joints and connective tissues [1]. Over the past decades, significant improvements in the treatment of RA have been accomplished. However, patients still experience impairments in their daily life [2]. One way to measure the impairments from the patient perspective is through patient-reported outcome measures (PROMs), mainly used for research purposes [3].

The shift towards a more patient-centered healthcare system enhances the role of the patient in daily practice regarding treatment decisions and subsequent outcomes [4]. Therefore, PROMs are considered to be central components within Value-Based Health Care (VBHC), as PROMs reflect outcomes from the patient perspective, aiming to improve the perceived value [5, 6]. Patient involvement within the VBHC strategy also induces the alignment of patients with respect to the process of integral healthcare delivery and therefore on the denominator of Porters' value equation, which defines patient value as health outcomes divided by costs [4].

Mapping patient pathways or so-called journeys in a systematic manner, provides insight in the value added by as well as the (in)efficiencies related to the provision of care of each activity, may reduce practice variation and on the other hand promote personalized care at the right place [7]. Several methods and frameworks to design patient journeys are available [7]. However, most methods lack a direct link to the relevant outcomes, e.g. clinical and/or PROMs, as a measure of the quality of care besides the healthcare costs involved. Within VBHC, the Care Delivery Value Chain (CDVC) was developed as a framework to facilitate the construction of patient journeys, encompassing patient relevant activities for a given medical condition [6]. Furthermore, the patient journey can be exploited in co-creation with patients to redesign the care cycle to improve the quality of the care provided as well as identifying the proper time horizon to analyze (patient reported) outcomes [7, 8]. Also, identified inefficiencies can be resolved to enhance value creation. The CDVC approach will be exploited to assess the organization of RA care at Maasstad hospital. The objective of this study is to define the CDVC and to establish a detailed process map of the care delivered within the standardized pathway criteria, i.e. RA patients following the regular patient journey. Areas of value creation will be identified based on the structured and practical mapping of the RA care cycle in collaboration with patients and the medical treatment team.

Methods

Study design and data collection

A mixed method research design was followed to assess the CDVC and map the activities concerning the patient journey. Quantitative data concerning the CDVC were collected from electronic health records. The Dutch Healthcare Authority requires medical personnel to register the performed care activities per patient and therefore the electronic health records contain detailed information on the healthcare procedures carried out within the patient journey.

Study population and setting

Quantitative

The prospective (open) cohort research was conducted in a real-life cohort of RA patients at the rheumatology department of Maasstad Hospital a top-clinical research hospital in Rotterdam, the Netherlands, from 2014 onwards. The inclusion criterion for the study was a RA diagnosis determined by a rheumatologist. Currently, over 3,100 diagnosed patients yearly medical care for RA in Maasstad hospital. Gender, disease duration and age at diagnosis were analyzed in the study population. StataSE version 15 was used for the descriptive statistics.

Qualitative

The qualitative data including time frames and division of labor were obtained from in-depth interviews with the staff members of the rheumatology department. Over 40 people work in the department, of which 12 are rheumatologist. The department functions as a training institute for rheumatologists and research is a high priority. The catchment area of the hospital is the fourth largest in the country.

The study started with a process map for the rheumatology department dating from 2014, which was updated by conducting interviews with the rheumatology medical team (supplementary file 1). The inclusion criterion of the cohort applied, was that one staff member per employer group was allowed to give unstructured feedback on the process map. Revisions were incorporated and the updated process map was sent to the treatment team of rheumatologists. A delegation of the rheumatologists validated the full care cycle by means of a single focus group. The researcher presented the patient journey including all activities, resources and timeframes in a chronological manner. An unstructured methodology was chosen, allowing medical staff members to raise any question regarding the RA care cycle.

2

Co-creation with patients

In 2016 a patient advisory board was initiated at the Maasstad hospital and in 2022 the board comprises of approximately 70 inflammatory rheumatic disease (IRD) patients of which around half suffer from RA. The purpose of the panel is multifold as Maasstad hospital strives to achieve active patient engagement and participation. Depending on the purpose of a gathering, events and meetings are held to inform, consult, ask for advice, co-create, co-decide with or stimulate self-management by IRD patients. For study purposes the RA care cycle was presented to the patient advisory board to evaluate and co-create the delivery of RA care from the patient perspective.

Outcomes

Care delivery value chain and process map

The CDVC was established by applying the method of Porter et al. to map and evaluate the process of care delivery [6]. The CDVC describes the main activities within the patient journey as well as the process flow and organization of the care delivery cycle. In the CDVC, inter- and intrapersonal communication between medical personnel are not incorporated. A detailed process map, including all activities, of the patient journey was constructed with the CDVC phases serving as a base. As part of the Dutch healthcare system, registration of the activities is required to properly reimburse the provided care [9]. The Dutch reimbursement system comprises over four thousand reimbursement codes, allowing for a detailed analysis of the individual care activities [9]. The process map gives an overview of the trajectory patients must follow regarding the activities as part of their RA treatment. Non-reimbursed activities such as registration of patients at the reception desk, were identified via interviews. The clustering in the CDVC was based on the output of the interviews with both the staff and the patients.

Improvement and optimization of the RA care cycle

Through the mapping of the CDVC we aimed to identify value improvements opportunities or areas concerning the optimization of the care delivery. The focus of the identification of improvement actions is mainly organizing care in alignment with the demands of patients suffering from a chronic disease.

Patient engagement

To examine the level of patient participation in the care delivery process, the framework of Carman et al. was applied [8]. In that framework three levels of engagement are distinguished: direct care, organizational design and governance and policy making [8]. The latter level of patient engagement is, however, not applicable for this study. For each level of engagement, the extent of patient participation is either on basis of consultation, involvement or partnership and shared leadership, i.e. the continuum of engagement [8].

Data visualization

Microsoft Visio (version 2016) was used to visualize the care cycle of RA patients [10]. Flowcharts consist of swimming lanes, delineating the total cycle in sub processes. Swimming lanes are either horizontal or vertical arranged. With respect to the RA patient journey, horizontal swimming lanes are used to distinguish the staff members and resources essential for the individual process steps. Staff members are denoted on the vertical axis in the margin of the swimming lanes. The patient journey starts in the upper-left corner of the flowchart. The representation of the flowchart symbols are described in the results section.

Results

Population characteristics

As of 2021, the Maasstad hospital RA population comprised of 3141 patients of which 71.5% female. The mean age at diagnosis is 57.7 (SD=15.0) and on average the disease duration is 7.8 years (SD=4.6).

Care delivery value chain

In figure 1 the CDVC for RA patients is displayed. Per element of the CDVC, the main activities with respect to the treatment of RA patients are summarized. The CDVC distinguishes eight different elements concerning the mapping of a care cycle, of which the bottom five describe the phases relevant for the actual process mapping. Informing and engaging, measuring and assessing occurs throughout the phases of the CDVC. In the following sections, these phases and the corresponding activities are described in detail for the RA care cycle.

Figure 1. Care delivery value chain of rheumatoid arthritis patients

| Informing and engaging Education on drugs, education on disease and treatment | | | | | | |
|---|--|---|--|---|--|--|
| Measurements Measurements Patient-reporte assessment | g : length, weight a d Outcome Meas | ind blood pressure. surements and Dise | Imaging, laborat ase Activity Scor | ory tests, e (DAS) | | |
| Accessing Outpatient visit | s, laboratory visit | ts, radiology visits a | ind telecare (rem | ote care) | | |
| Diagnosing - Referral - Lst consult (rheumatokgist) Diagnostiss: imaging & laboratory - Consult: diagnosis - Education | Preparing - Assessments & measuresments - DAS assessment - Drug verification | Intervening - Disgnostics: biboratory tests - DAS assessment - Consult: drug course - Consultations - Drug verification | Recovering - Diagnostics: laboratory tests - D4S assessment - Consultations - Drug verification | Monitoring/managing - Dignostics: bboratory tests - DAS assessment - Drug verification - Consultations | | |

Informing and engaging, measuring and accessing

Informing and engaging

Education concerning the disease (e.g. shared-decision making) and the pharmacological treatment, as part of the informing and engaging process, is mainly provided in the first half year after the diagnosis (Table 1). After this initial phase, the treatment team provides education on request or when needed, for example when patients switch between drugs or taper medication intake. Apart from education provided by staff members, patients can actively engage by virtue of the ReumaWeb application, which is a self-management tool designed to provide remote coaching consisting of information and exercises on how to alleviate RA-related complaints [11].

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| Phase | # Of activities | Informing and engaging | Measuring | Accessing |
|---|--------------------|---|---|--|
| Diagnosing | 11 | Disease education | Screening disease activity, i.e. blood tests and x-rays | GP practice Outpatient rheumatology department Laboratory Radiology |
| Preparing | 5 | Education, i.e. drugs and non- drug Drug information | Disease activity measurement (DAS28) Baseline measures (length, weight blood pressure) | Outpatient rheumatology department Hospital pharmacy |
| Intervening | 11 | PROMs education and information Education, i.e. drug side effects | Screening disease activity, i.e. blood tests | Laboratory Outpatient rheumatology department Hospital pharmacy |
| Recovering (from symptoms) | 12 | Upon request or when needed ReumaWeb app® | Screening disease activity, i.e. blood tests Disease activity measurement (DAS28) | Laboratory Outpatient rheumatology department Hospital pharmacy |
| (Life-long) monitoring and managing | 7 | Upon request: education, i.e. drugs ReumaWeb app® | Disease activity measurement, i.e. DAS28 Patient Reported Outcome Measures (DQRA, FACIT, EQ-5D, HAQ-DI, POP-66, RAID, WPAI)* | Laboratory Outpatient rheumatology department Remote consultations Hospital pharmacy |
| Total | 46 | | | |

Table 1. Informing and engaging, measuring and accessing within Rheumatoid Arthritis care cycle

*DQRA Dutch quality registry rheumatoid arthritis, FACIT Functional assessment of chronic illness therapy fatigue scale, HAQ-DI Health assessment questionnaire disability index, RAID Rheumatoid arthritis impact of disease, WPAI Work productivity and activity impairment

Measuring

At the beginning of the care cycle, the patient's medical condition is assessed by measuring the disease activity. Towards the monitoring phase PROMs are becoming increasingly important. In addition to the usual clinical examinations, PROMs are gradually integrated as part of the daily clinical practice to represent the outcomes that matters most to RA patients and help to determine treatment goals. For this purpose, the ICHOM standard set for inflammatory diseases was implemented measuring various health domains [12]. Prior to implementing the PROMs at the department, four panel meetings with patients from the patient advisory board were held to discuss and evaluate the PROMs questionnaires and the tool visualizing the results [13]. In addition to RA-related health outcomes, social participation of the patients is also observed by examining productivity at work and mental well-being. Patient's evaluation of the care

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delivered is integrated in the PROMs amid the DQRA, a quality register for care facilities treating Rheumatoid Arthritis in the Netherlands. As the DQRA measures patients' perceptions of their care delivery experience, the DQRA is considered as a PREM (Patient Reported Experience Measure).

Accessing

Sites of care delivery are most often the various departments in the hospital as displayed in Table 1.

Patient journey RA

The detailed process map considering the RA population, i.e. patients suffering solely from RA, is presented in the supplementary materials (supplementary file 2). On average, patients visit the rheumatologist and the doctors assistant six times and the physician assistant/nurse practitioner three times in the first year. Visits to the rheumatology nurse total three and counter employees perform 15 activities within the patient journey. Drugs are picked up at the hospital pharmacy during (six of) the onsite visits. Diagnostics, i.e. imaging and blood testing, is conducted eight times.

Diagnosing

The diagnostic phase of the RA care cycle contains 11 distinctive steps, involving four different healthcare professionals. The degree of variation is largest in the diagnosing phase with respect to the staff members patients consult and the succession of onsite appointments. The focus of this phase is to properly diagnose patients within three weeks, but as timely as possible. Based on the diagnosis, patient-tailored education is provided.

Preparing

The preparing phase consists of five steps. Since this phase commences immediately after the diagnosis, patients are not compelled to visit the outpatient department solely for the preparing phase. Informing patients on the nature of their illness is of importance as it may affect the treatment responsiveness positively as a result of therapy compliance. Therefore, the target of the preparing phase is to increase patient activation through supporting and engaging patients in the treatment by informing.

Intervening

The intervention phase follows the preparation phase. A total of 11 activities were distinguished in the intervening phase; the process is divided in two parts, taking place with an interval of approximately six weeks. The corresponding steps in the parts are
similar, however the consultation with the rheumatologist is replaced by a consultation with the physician assistant or the nurse practitioner. The focus of the initial intervention phase in RA is obtaining a suitable treatment setting for patients, i.e. treat-to-target. Treat-to-target is setting a clinical target such as low disease activity or remission, choosing the treatment through shared-decision making and accomplishing the clinical target [14]. Assessment of the determined target is conducted, in consultation with the patient, via for example the disease activity score, the primary clinical outcome measure advise by international guidelines [15].

Recovering (from symptoms)

In general, the recovery (from symptoms) phase begins three months subsequent to the intervention and consists of 11 steps. Activities in the recovery phase are comparable to the activities in the intervention phase. However, the focus is on achieving the (agreed) clinical target. And, if possible, tapering of the drugs are pursued without causing a flare-up of the disease. Since RA is a chronic disease and full recovery is unattainable, the designation is adjusted to the recovery or stabilization from symptoms. During recovery, the disease activity of the patient is expected to decrease as a result of effective treatment.

(Life-long) monitoring and managing

Monitoring and managing starts on average after 48 weeks of treatment and continues thereafter. In case of inflammation of the joints or a high disease activity, due to the nature of the illness, patients will deviate from the time span illustrated in the patient journey. With respect to the conventional care process, the steps for patients are in line with the recovery phase. The frequency of consultations in the monitoring phase varies between the different staff members and is dependent on whether a patient experiences flare episodes. After one year the DAS assessment and blood drawing takes place approximately every 12 to 24 weeks, depending on the level of disease activity of a patient. In total, monitoring and managing regular care counts seven successive steps.

Validation process map

Once the detailed process map was charted, the patient journey was presented and distributed amongst the members of the rheumatology patient advisory board. Patients were encouraged to provide comments on the patient journey either during the meeting or via e-mail. At the board meeting, the patients confirmed the outlined steps and therefore the validation did not lead to significant adjustments within the process map. Furthermore, no e-mails were received concerning comments to the patient journey. With respect to the validation amongst the medical staff of the rheumatology

department, a few comments were made concerning the stated time frames and were discussed during the presentation of the patient journey. Therefore, one adjustment was made with respect to the time frame within the diagnostics stage.

Improvement and optimization of the care cycle

Patient engagement

To improve the care delivery, patients are stimulated to actively participate in the evaluation of the patient journey. Regarding the first level, i.e. direct care, patient's engagement is at the higher end of the engagement continuum. This is due to the fact that treatment is based on patient preferences through shared-decision making and PROMs assessment. The intermediate level, involving patients, applies to organizational design and governance (second level). Perspectives of the patients in the patient advisory board are considered in the design and evaluation of the care process as described below.

Process level

After analyzing the patient journey by means of the CDVC, several improvement areas were mentioned during the interviews and meetings with patient partners and caregivers. Pertaining to the accessing phase, it was noticed that the care sites are primarily the hospital departments. With the recent development of shifting healthcare beyond the hospital walls and the COVID-19 pandemic, a telemonitoring track was initiated by Maasstad hospital to expand the access to healthcare and to reduce the burden on patients. Despite the fact that patients can have electronic consultations, it is still necessary to visit the hospital for blood drawing. By facilitating drawing blood at home, the full monitoring phase will be shifted from the hospital site to the home of patients, reducing transportation costs and time burden on patients. Patients participating in the patient panel indicated their preference of blood drawing at home as opposed to the hospital. The suggested telemonitoring track was also discussed and approved by the staff of the rheumatology department. In the near future, in addition to the performed e-consultations, patients will be able to carry out blood drawing at home.

Outcome level

An outcome level improvement was identified to support the performance of DAS assessments at home as part of the telemonitoring track. The standard DAS assessment will be substituted or complemented by a patient-reported DAS assessment in the telecare process. As a result, patients are able to fill out the DAS assessment remotely and are not required to visit the hospital. Another outcome improvement action was related to the measuring phase of the CDVC. To measure PRO's in a consistent

manner, the improvement action focuses on a frequent measurement. Therefore, patients are requested to fill out the PROMs every six months in line with the ICHOM recommendations. A baseline measure will be performed in the preparation phase. Thereafter, patients will be requested to complete the PROMs semi-annually.

Structure level

Concerning the improvements with respect to the organization of the RA healthcare delivery, another improvement area was identified. Applying PROMs as guidance to arrange the recurrence of appointments in the monitoring and managing phase. Thereby enhancing patient involvement and patient value since care delivery is customized to individual patients. The timing and organization concerning the education given by rheumatology nurses is also investigated. In the former situation, education was solely provided after the diagnosis in the preparing phase. As a result of a focus group with 16 patients, a second consultation with the rheumatology nurse is added to the patient journey to gain more knowledge at the follow-up in the intervening phase.

Discussion

The results of the study demonstrated that by applying a mixed method design the CDVC is a useful method to structure and gain insight in the real-life care delivery cycle of RA patients. Despite the clustering in the CDVC process steps, many of the performed procedures in the steps are similar. Yet, the focus of the different process steps in the care cycle vary. The focus shifts, from predominantly a treat-to-target strategy and examining disease activity at the start of the cycle, towards improvement on tapering medication and managing the disease on a more personalized basis through PROMs. Although the patient's-maintained functioning, quality of care and the patient's evaluation of the received care are monitored, reflection of the CDVC and patient journey resulted in several improvements on outcome and process level.

The CDVC is considered as the basis for the integration of VBHC in healthcare delivery. By applying this methodology, we have gained knowledge concerning the arsenal of the delivered chronic care services, ultimately leading to personalized value of RA treatment (personalized outcomes related to personalized journey cost). A perspective facilitating to complement and synergize the classical focus of evidence based medicine and towards improving value by reorganization of the care delivery on the meso and macro level, where besides clinical evidence also evidence on patient report outcomes and healthcare cost are included [16][17]. Ultimately, the goal is to provide personalized care on a micro

level by engaging patients through consulting, involvement and evaluation of the care delivery process [8]. Moreover, the creation of the process map and mixed methodology of the RA patient journey allows for uniform analyzes and can therefore be considered as a blueprint for other (chronic) illnesses.

The self-reported DAS, one of the improvement actions, is a next step in the treatment of RA patients remotely. However, a possible challenge could be the level of reproducibility of the self-reported DAS to a clinical reported DAS. A systematic review conducted by Rampes et al. demonstrated that both the total joint count (TJC) and small joint count (SJC) were reliable when performed by patients [18]. In a similar study, the CDVC was created for HIV/AIDS in Togo, enabling the identification of quality improvements with respect to the chronic care delivery for pediatric HIV and AIDS [19]. However, a distinction was the fact that the care delivery consists of many more facets in comparison to the RA care cycle. Moreover, in this study, the CVDC was utilized to construct a detailed process map and to determine the level of patient engagement in the care delivery with the framework of Carman et al. to investigate the level of patient engagement in RA care delivery has not been carried out before. The latter is of importance from the VBHC perspective, as the input of patients and patient engagement should be enhanced.

In prior research the RA patient journey was developed by conducting interviews with patients and combining the results of the interviews with process mapping [20]. The focus of the study by Oliver et al. was predominantly on depicting patient experiences concerning the care delivery. A validated graphical representation of the process was however lacking. Moreover, implications for improvements were mainly aimed the access to rheumatology care. Hence, in this study an illustration of the patient journey is given on basis of the CDVC and the improvement actions are not confined to specific phases of the care cycle. Another methodology, as mentioned by van Weert and Hazelzet (2020) [21], is presenting the patient journey through so-called "metro mapping", in which the patient journey is depicted as a metro line [22]. To date the majority of metro mapping is performed within elective (oncology) disciplines rather than chronic care and the metro lines method restricts the application of loops, returning to a previous activity, within the care cycle. Furthermore, the metro mapping of care cycle is conducted by educated service designers, making it a less accessible method compared with the CDVC, which can be performed by anyone of the organization [22].

An additional strength is related to the engagement of healthcare providers in the process of mapping, improving the efficiency of the delivery of care and identifying practice variation as processes were discussed and, in the end, formalized. Moreover, the established CDVC and detailed process map were presented and discussed with the patient panel. Defining improvement actions and the patient care implementation of telecare in the CDVC to increase patient value are additional strengths of the research. A last strength is attributed to the financial organization of the Dutch healthcare system where care activities are broadly registered and information with respect to care activities within patient journey are easily accessible. Therefore, activities and costs can also be easily matched.

A limitation of the study is the fact that the patient journey was limited to RA patients treated in the hospital silo (secondary care). The integral process, i.e., primary care and tertiary care, could not be included in the mapping of the patient journey due to the pillarization of the Dutch healthcare system. However, the majority of the outcome and procedures is carried out at secondary care institutions and therefore, insight in this part of the patient journey is of great relevance. A second limitation of the study concerns the process steps described in the CDVC. Allocating activities to the phases of the CDVC is to a certain extent subjective and can therefore be interpreted differently, potentially complicating the CDVC benchmark. However, the guidelines with respect to the care of RA are nationally established, Moreover, as the CDVC was validated by the medical staff and the patient advisory board the potential bias is limited. The validation of the care cycle was limited to an internal validation, which can be considered a final limitation as it reduces the generalization of the results. a next step is to externally validate the CVDC and process map within other hospitals in- and outside of the Netherlands.

Conclusion and future perspectives

The aim of the study was to define the patient journey for the standardized care delivery in single-morbid RA patients. However, in the remaining, around one-fifth of the patients, the pathway may deviate due to patient's characteristics e.g. multimorbidities, preferences and unforeseen circumstances. Therefore, further investigation is required to address challenges such as the occurrence of multimorbid and complex patient populations in the care cycle. As a next step, allocation of healthcare costs to the activities within the CDVC, will provide insight into the (integral) healthcare costs associated with different patient journeys. Obtaining and allocating the related costs and time of the RA CDVC allows for a sophisticated analysis of the care cycle, contributing to the transition to a value-based healthcare system. In the end, the goal is to evaluate outcomes in relation to costs on a patient level. In conclusion, establishing the CDVC and process map for RA provided detailed information regarding the patient journey of RA patients in an effective and value attaining manner. The study provided transparency in and standardized the various processes, activities and involved staff members in the delivery of RA care. As a result, improvement areas were easily identified, and the implementation of patient telecare were facilitated. 2

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Appendix



Supplementary file 1. Process map rheumatology department 2014

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Supplementary file 2. Detailed process map (updated)

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Chapter 3

Dealing with Time Estimates in Hospital Cost Accounting

Koster F, Kok MR, van der Kooij J, Waverijn G, Weel-Koenders AEAM and Lopes Barreto D

Pharmacoecon Open. 2023 Jul; 7(4): 593–603, doi: 10.1007/s41669-023-00413-2

Abstract

Time-driven activity-based costing (TDABC) can support value-based healthcare (VBHC) programs by providing insights into the actual relationships between time spent by the medical staff and the costs associated with specific care cycles. However, the robustness of time estimates (time variation) as well as the effort required to obtain these estimates are major challenges of the TDABC methodology, given the heterogeneity in patients' needs and the presence of (multi)morbidity. To allow for the variation in time estimates in an efficient manner, this study uses fuzzy logic (FL) to estimate the TDABC model parameters (FL-TDABC).

A standardized care path was used to calculate the annual costs (per patient) and cost drivers of the Rheumatoid arthritis (RA) care cycle following the FL-TDABC methodology. Cost information (2018) was derived from hospital reports concerning financial, human resource and business intelligence data from a Dutch top clinical research hospital, Maasstad Hospital. Time estimates of procedures were obtained by interviewing the medical staff and relevant care activities were extracted from electronic health records. For analytical and validation purposes, FL-TDABC estimates were compared with TDABC and ABC cost estimates.

The RA care cycle annual costs totaled €1497 per patient (2018 prices) based on the FL-TDABC methodology. Maximum RA cycle costs (€1684) were some 22% higher than minimum costs (€1317) observed from FL-TDABC. Cost drivers explaining the cost variation are predominantly the number of consultations with rheumatologists and pharmacy costs related to RA. Based on TDABC and ABC, annual costs per patient were €1609 and €1604, respectively.

The FL-TDABC methodology offers a more precise and efficient estimate of care cycle costs, allowing for the subjective (fuzzy) nature of healthcare time estimates made by the medical staff. As a result, the FL-TDABC provides insight into the practice variation, and hence it can promote the transition from a volume-based system to a VBHC system.

Introduction

Due to a lack of transparency in healthcare expenditures, governments and decision makers are inhibited from adequately containing the growth of costs [1]. Over the years, various management tools have been proposed as possible solutions for the high level and rapidly increasing expenses. However, strategies such as lean and six sigma cannot hold healthcare practitioners appropriately accountable for the quality and costs of care delivery due to the lack of time and resources to implement those tools [2]. Another drawback of lean and six sigma is the one-dimensional focus on processes, instead of the preferred holistic perspective [3].

Policy makers should make informed decisions with respect to the allocation of resources within healthcare, which requires transparency of the costs associated with the treatments provided [4]. Cost information can offer managerial value if costs can be linked to the factors that drive them, such as individual procedures in a care cycle. Several cost allocation methods have been developed to provide insight into the drivers of healthcare costs on a micro level [5].

The practice of cost accounting and allocation in healthcare has evolved over time, leading to, inter alia, the development of time-driven activity-based costing (TDABC). TDABC was introduced by the Harvard Business School in 2003, as an elaboration of the traditional activity-based costing (ABC) approach [6]. The TDABC methodology makes use of two variables, namely the required time commitment by a resource and the capacity cost rate (CCR) of that resource [6]. From a value-based healthcare (VBHC) perspective, calculating the costs of care delivery, i.e., the activities performed regarding the treatment of a disease, is an essential component [7]. Therefore, TDABC has been proposed as the principal cost accounting method within the VBHC framework [7].

A drawback of traditional ABC, which underlies the current Dutch reimbursement system within healthcare, is the fact that the time spent by medical staff to perform certain treatments is based on point estimates. In the case of the Dutch healthcare reimbursement system, these point estimates are not disclosed (black box): healthcare cost reimbursements comprise a lump sum for every standardized diagnosis treatment combination (DTC), of which there are around 4000. However, information about the time factor (time spent per resource) is of great importance to achieve transparency in costs, as patients experience (multi)morbid conditions that lead to variations in the duration of consultations with healthcare professionals. Especially in an era of shared decision making, time spent by medical staff is a key driver of healthcare costs. As

TDABC also uses point estimates for the time required to perform medical procedures, TDABC can also be assumed to be biased [8]. To deal with the variation in time estimates regarding healthcare activities, this study combines TDABC with the fuzzy logic (FL) theory, which uses pooled time estimates instead of point estimates. The use of FL is based on the observation that medical and healthcare data tends to be subjective (fuzzy). FL-TDABC has shown to be an effective strategy in healthcare and other sectors as the full spectrum of possible conditions from a best- to worst-case scenario can be taken into account [8–10]. Furthermore, implementing and maintaining a TDABC model is a labor-intensive and time-consuming procedure, predominantly carried out by researchers [11].

The aim of this study was to apply the FL-TDABC methodology to the Rheumatoid Arthritis (RA) standardized care cycle in Maasstad Hospital, to estimate the treatment costs per patient, considering the variation in time spent by medical staff to perform activities within that care cycle. Variation in the actual time committed by the medical staff per patient is caused by the heterogeneity of the RA patient population in terms of, inter alia, the complexity of their medical status, i.e., experiencing multimorbidities. FL-TDABC is considered to be a less demanding cost allocation method than traditional ABC or TDABC for the determination of the time required to perform healthcare activities. Furthermore, FL-TDABC can provide information regarding the cost drivers of the standardized RA treatment cycle that can be used in the decision making concerning healthcare resource utilization and allocation. The current healthcare cost reimbursement system in The Netherlands, which is based on the ABC methodology, does not provide that information, as the time estimates underlying the calculations have not been disclosed.

Methods

Study Design and Population

For this TDABC study, the standardized care path for RA patients, developed at the Rheumatology and Clinical Immunology Department of Maasstad Hospital, a top clinical teaching and research hospital in Rotterdam, The Netherlands, was used [12]. For valuation purposes, 2018 cost estimates were used since 2020 and 2021 prices were affected by the coronavirus disease 2019 (COVID-19) pandemic and therefore did not represent the actual, competitive price. Identification of the patient's medical condition corresponds with step one of the TDABC framework [13]. RA is a chronic disease causing inflammation of joints and tissues [14]. RA patients require lifelong treatment with the

goal of diminishing the disease activity in patients, mainly achieved by administering disease-modifying anti-rheumatic drugs. Future treatment within the RA patient population will focus on the cost-effective and patient-centered delivery of care [15].

Data collection

To obtain the research data, a mixture of quantitative and qualitative methodologies were applied. Quantitative data were collected via hospital reports concerning financial and human resource data. With respect to the financial data, the annual salaries of the medical staff and the direct and indirect costs of the performed activities were assessed [16]. Human resource data concerned the medical staff capacity, in full-time equivalents (FTEs) and average work days per week. Data concerning time estimates were acquired by interviewing the staff, and care activities were obtained from electronic health records in 2021. A total of six counter employees, five rheumatologists, four doctors' assistants, three physician assistants (or physician associate)/nurse practitioners, and three rheumatology nurses (i.e. nurses specialized in rheumatology) participated in the questionnaires. A physician assistant is a nurse or paramedic who has completed advanced training and who can independently perform complex medical tasks and actions, otherwise performed by a rheumatologist. As part of the ongoing JOINT Evaluation study, evaluation of the (clinical) data reported at the Rheumatology Department is allowed by the hospital board (MEC number: T2016-76).

Fuzzy Logic Time-Driven Activity-Based Costing (FL-TDABC)

The seven-step approach established by Kaplan and Porter [13] was followed to systematically gain insight into the costs of the standardized care cycle of RA patients (Fig. 1) [13]. The first three steps served as the basis for the present study. In brief, based on the medical condition defined (step 1), the care delivery value chain (CDVC) (step 2) and detailed process map for RA patients (step 3) receiving treatment in Maasstad Hospital were determined [12]. The emphasis of this study was to investigate and elaborate on steps four to seven of the approach, as shown below (Fig. 1).



Figure 1. Time-driven activity-based costing framework in healthcare

The TDABC methodology was modified by including FL in the approach (Fig.1) [13]. Cost prices were calculated by allocation of resources to cost centers using two variables: the CCR of a resource and the time commitment by that resource [17]. The CCR is the total of all costs required to supply a resource, such as a consultation or a laboratory analysis, divided by the time required to supply that resource by the employees [17]. Since RA is a chronic disease, the duration of the care cycle was set at one year as recommended by the Harvard Business School [13]. The healthcare providers involved or other resources required and the place of care delivery were denoted per activity (i.e. procedure) that patients undergo [12].

Time Estimation of the Activities in the Process Map (Step 4)

Fuzzy Logic

The FL theory was developed by Zadeh to improve the logic of fuzzy data sets, i.e., data sets with imprecise information [8][18]. Imprecise information in healthcare is predominantly the result of the variability of the time spent on procedures. FL allows managers and policy makers to make decisions based on time estimates that are closer to clinical practice, which has a positive effect on the organization of care. In this study, the methodology of the triangular fuzzy numbers (TFNs) is applied, where TFNs represent a category of fuzzy numbers reducing the ambiguity of outcomes, specifically with respect to the variation in time commitment required by the

medical procedures within the diagnosis-related groups (DRGs) between caregivers [8]. TFNs are composed of three parameters (Eq. 1): the smallest possible value (a), the most promising value (b), and the largest possible value (c) [8]. The most promising value is defined as the modal value or the most frequently occurring value. Equation 1 shows the function underlying the FL estimate, which is applied to calculate the average costs of the activities (x*). As opposed to TDABC, FL accounts for the variation in possible outcomes due to the fact that the calculation is based on three outcome parameters. Within TDABC, the labor-intensive stopwatch timing is predominantly used to obtain time stamps of procedures, which is a more subjective and time-consuming method.

Equation 1. Center of gravity—fuzzy logic estimate

$$x^* = (a+b+c)/3$$

In TDABC, for generic, low-cost and brief procedures, standard times were used. As standard times were unavailable for X-rays of the hands, feet and lungs, as well as for laboratory tests, the time to perform imaging and laboratory tests could therefore not be included in the cost calculations. For the complex activities within the RA care cycle, time estimates were obtained via interviews or direct observations. In the FL-TDABC analysis, the time spent on complex and expensive procedures was estimated according to the FL method. Through questionnaires, rheumatology staff were asked to estimate the time they spent on activities such as consultations, patient registration and disease activity score (DAS) assessments. The smallest possible value, the most promising value, and the largest possible value in terms of the time required to perform these activities were assessed. The most promising value indicates the most frequently occurring time estimate as a result of inquiry among the medical staff.

Cost estimation of supplying patient care resources (step 5)

Estimates of the cost of supplying patient care resources were obtained through assessment of the direct and indirect costs related to the treatment of the medical condition RA [13]. Direct costs included all activities immediately associated with the care delivery, such as consultations, materials, laboratory processes and medication. Indirect costs consisted of general costs such as IT, housing and electricity, and these costs were allocated by the finance department via allocation keys. Honorarium costs were separated as these costs are used to calculate the FL-TDABC estimates. Laboratory costs consisted of the direct, indirect and honorarium costs of the frequently conducted blood tests, with exception of the laboratory costs in the diagnosis stage. Direct, indirect and honorarium costs concerning the x-rays were based on standard prices. Pharmaceutical costs were allocated to the drug verification procedure comprising the pharmacy assistant. Medication use by patients is accounted for in the calculation of the pharmacy costs. These direct, indirect and honorarium costs, if available, were allocated to the activities in the process map.

Estimation of the capacity of patient-specific resources and calculation of the capacity cost rate (step 6)

The CCR was calculated by dividing the total costs per patient-specific resource by the practical capacity available. Practical CCRs were expressed as the cost per minute for the deployment of medical staff members. The practical capacity was derived by extracting the actual care delivery hours, excluding time spent on meetings, education, administration, holidays, sick leave, and breaks. Annual salary costs, including an employer mark-up of 30% (for social security and pension premiums), and the FTE per function were considered in calculating the costs per minute of the medical staff.

Total cost calculation of the rheumatoid arthritis care cycle (step 7)

The final step of the TDABC model for healthcare is calculating the costs of the cycle of care for a patient [13]. The time spent per resource was multiplied by the capacity cost rate. Costs were aggregated to obtain the costs of the care delivery cycle for RA patients. The CCR per resource multiplied by the duration of the activity (in minutes) per phase, gives the costs of every process step [13]. The FL-TDABC total costs of treating a RA patient was calculated as the unweighted average of the total costs comprising all process steps for the smallest possible, most promising and largest possible values [13]. To analyze the variability in costs, the smallest, most promising and largest values are also displayed in the Results section.

Time-Driven Activity-Based Costing

To compare the cost estimates with the ABC and TDABC estimates, in TDABC, standard times are used for the TDABC method instead of the FL time inputs. Calculation of the TDABC value is based on the methodology as defined by Kaplan and Porter [13]. The CCR and the time commitment were applied to calculate the prices per activity or resource with respect to the honorarium costs [17]. Direct and indirect costs do not vary between the FL-TDABC and TDABC, apart from the honorarium costs. Therefore, instead of applying FL time estimates as in FL-TDABC, standard time point estimates were considered in TDABC. Standard times are nationally determined with respect to consultations with rheumatologists [19]. The standard time for the first consultation of an RA patient is 50 min, the consultations within 120 days after diagnosis are set at 40 min, and follow-up consultations are given 15 min. Consultations with physician

assistants and nurse practitioners are set at 15 min in Maasstad Hospital. No standard times are available for other staff members (e.g. rheumatology nurses, doctor's assistants and counter employees) and therefore the mean value of the FL estimate was applied as a reference to estimate the TDABC care cycle costs.

Activity-Based Costing (ABC)

Comparisons between the different cost accounting methods were also made with respect to the traditional ABC method. ABC costs are estimated by calculating the average DTC, in Dutch (*Diagnose Behandeling Combinaties* [DBC]), costs per RA patient in the first year of RA treatment. The ABC value was calculated by dividing the sum of the DTC costs by the number of patients in the RA patient population (2837 patients were included in the cost calculation). Hence, this calculation is not based on the standardized care path. The Dutch DTC codes comprise the registration, diagnosis, treatment and billing codes with respect to diseases treated in specialized medical care [20]. Cost reimbursements based on DTC codes are calculated by a grouper, an automated cost allocation system, centrally organized in The Netherlands with mandatory validation for reimbursement (see Appendix) [20]. Based on the nature and number of care activities provided to the patient, the most appropriate DTC product is selected by the system and a price calculated and communicated to the health insurer(s) [20]. In The Netherlands, over 4000 DTC codes exist for the specialized medical care provided in hospitals [20].

Statistical Analysis

To test the significance of the difference between the current ABC (i.e. DTC) model versus the FL-TDABC model and the TDABC model using standard times, two one sample *t*-tests were conducted. The two values concerning FL-TDABC and TDABC, obtained from the standardized, average care path and therefore considered as average total costs, were compared with the mean costs of the ABC (i.e. DTC) estimate as mentioned in the previous paragraph. The FL-TDABC and TDABC values are compared with the ABC (i.e. DTC) value, since the ABC method underlies the current costing application in the Dutch healthcare system. Because samples sizes were large and therefore the central limit theorem holds, parametric tests were conducted. Stata/SE 15 for Windows (StataCorp LLC, College Station, TX, USA) was used to perform the statistical analysis and an alpha level of 5% was considered as statistically significant.

Results

Study Population

The study results were based on the mapped care path of RA patients diagnosed by a rheumatologist in the Maasstad Hospital, Rotterdam, within the CDVC [12]. More than 2800 patients received medical care for RA in Maasstad Hospital. The majority of the patient population was female (71.5%), and on average, patients were 57.7 years of age (standard deviation [SD] 15.0) when first diagnosed with RA by a rheumatologist.

Time Estimates

The CDVC and detailed process map presented in the Methods section were applied as the starting point for this study [12]. Time estimates per activity and resource are shown in Table 1. The FL methodology was adopted with respect to the complex and more timedependent healthcare activities, whereas for the remaining activities, standard time estimates were used. In absolute terms, the difference between the smallest and largest possible value for the time estimates (FL time) is the highest for the first consultation ($\Delta = 22$ min, range 19–41) and diagnosis consultation ($\Delta = 15$ min, range 10–25) with the rheumatologist and the education (e.g., instruction) activity of the rheumatology nurse ($\Delta = 15$ min, range 15–30). In relative terms, appointment scheduling by counter employees (82%) and the DAS assessment/PROMs activity of the doctor's assistant (67%) show the largest variances in the FL estimates.

| Care delivery value | | A | Standard time | FL time (min) | | |
|--|--|--------------------------|------------------------|---------------|----|----|
| chain phase | Provider/resource | Activity | (min) | Ts | Tm | ΤI |
| | Referrer (i.e. GP) | Consultation referrer | 10 | | | |
| | Referrer | Referral | NA | | | |
| | Counter | Registration | NA | 5 | 7 | 9 |
| | Rheumatologist | First consult | 50 | 19 | 29 | 41 |
| Diagnosing | Counter | Schedule appointments | NA | 2 | 5 | 11 |
| | Diagnostics | Laboratory | NA | | | |
| | Diagnostics | Imaging | NA | | | |
| | Counter | Check in | NA | 3 | 5 | 8 |
| | Rheumatologist | Consult: diagnosis | 40 | 10 | 15 | 25 |
| - · | Rheumatology nurse | Education/PROMS | NA | 30 | 38 | 45 |
| Preparing | Doctors' assistant | DAS assessment | NA | 5 | 8 | 12 |
| Care delivery value chain phase | Provider/resource | Activity | Standard time (min) | | | |
| | Pharmacy assistant | Drug verification | 15 | | | |
| | Counter | Schedule appointments | NA | 2 | 5 | 11 |
| | Counter | Check in | NA | 3 | 5 | 8 |
| | Diagnostics | Laboratory | NA | | | |
| | Doctors' assistant | DAS assessment/PROMS | NA | 3 | 6 | 9 |
| | Rheumatology nurse | Consult: drug course | NA | 15 | 26 | 30 |
| | Rheumatologist | Consult | 40 | 7 | 11 | 14 |
| | Pharmacy assistant | Drug verification | 15 | | | |
| Previder/resource Referrer (i.e. GP) Referrer Counter Rheumatologist Diagnostics Diagnostics Diagnostics Diagnostics Diagnostics Counter Rheumatologist Pharmacy assistar Counter Diagnostics Doctors' assistar Rheumatology n Rheumatology n Rheumato | Counter | Check in | NA | 3 | 5 | 8 |
| | Diagnostics | Laboratory | NA | | | |
| | Doctors' assistant | DAS assessment | NA | 5 | 8 | 12 |
| | Rheumatology nurse | Consult: drug course | NA | 15 | 26 | 30 |
| | Physician assistant/ nurse practitioner | Consult | 15 | 12 | 14 | 18 |
| | Counter | Schedule appointments | NA | 2 | 5 | 11 |
| | Counter | Check in | NA | 3 | 5 | 8 |
| | Diagnostics | Laboratory | NA | | | |
| | Doctors' assistant | DAS assessment/PROMS | NA | 3 | 6 | 9 |

Table 1. Fuzzy logic time estimations for the rheumatoid arthritis care cycle

3

| Care delivery value | Brovider (resource | Activity | Standard time | FL time (min) | | |
|------------------------------------|--|---|------------------------|---|----|----|
| chain phase | Flovider/lesource | Activity | (min) | Ts | Tm | TI |
| | Rheumatologist | Consult | 10 | 7 | 11 | 14 |
| | Pharmacy | Drug verification | 15 | | | |
| Decovering | Counter | Schedule appointments | NA | 2 | 5 | 11 |
| Recovering | Counter | Check in | NA | 3 | 5 | 8 |
| | Diagnostics | Laboratory | NA | | | |
| | Doctors' assistant | DAS assessment | NA | 5 | 8 | 12 |
| | Physician assistant/ nurse practitioner | Consult | 15 | 12 | 14 | 18 |
| | Laboratory | ActivityStandard time (min)FL tir TsConsult107Drug verification15Schedule appointmentsNA2Check inNA3LaboratoryNA5Consult1512Drug verification152Drug verification152Drug verification152Drug verification152Check inNA2Drug verification152Check inNA3LaboratoryNA3DAS assessment/PROMS1512Consult1512Consult1512Consult107ActivityStandard time (min)7Schedule appointmentsNA2 | | | | |
| | Counter | Schedule appointments | NA | time FL u Ts 7 2 3 5 12 2 3 3 3 12 3 12 7 time 2 | 5 | 11 |
| | Counter | Check in | NA | 3 | 5 | 8 |
| | Diagnostics | Laboratory | NA | | | |
| Monitoring and | Doctors' assistant | DAS assessment/PROMS | NA | 3 | 6 | 9 |
| managing | Pharmacy | Drug verification | 15 | | | |
| | Physician assistant/nurse practitioner | Consult | 15 | 12 | 14 | 18 |
| | Rheumatologist | Consult | 10 | 7 | 11 | 14 |
| Care delivery value chain phase | Provider/resource | Activity | Standard time (min) | | | |
| | Counter | Schedule appointments | NA | 2 | 5 | 11 |

FL fuzzy logic, Ts smallest possible value, Tm most promising value, Tl largest possible value, NA not available, GP general practitioner, PROMs patient-reported outcome measures, DAS disease activity score

Costs of the Care Cycle

The calculated capacity cost rates for the (medical) staff members are depicted in Table 2, which are used for calculating the total care costs. Regarding the first year of RA treatment, rheumatologists have the highest CCR (\notin 2.89 per minute), followed by the rheumatology nurse (\notin 1.41) and the physician assistant/nurse practitioner (\notin 1.24). Costs of a counter employee, doctors assistant and pharmacy assistant were less than \notin 1 per minute.

Table 2. Capacity cost rates per staff member

| Staff member function | CCR (€ per minute) |
|--|--------------------|
| Counter employee | 0.94 |
| Doctors' assistant | 0.77 |
| Pharmacy assistant | 0.36 |
| Physician assistant/nurse practitioner | 1.24 |
| Rheumatologist | 2.89 |
| Rheumatology nurse | 1.41 |

CCR: capacity cost rate

Given the CCRs, costs are calculated per CDVC step (Table 3). The total cost for treating a RA patient during the first year following diagnosis is calculated at €1497 using the FL-TDABC methodology. The diagnostics phase represents 32% of the total costs. Imaging (e.g. x-rays of the hands, feet and lungs), laboratory testing and the extensive time required for the first and second consultation with the rheumatologist account for the majority of the costs in the diagnostics phase of the CDVC. Within the CDVC, recovery and intervention account for nearly half of the costs due to the high frequency of followup visits within the first year, predominantly consultations with the more expensive rheumatologists and rheumatology nurses. Monitoring and managing and preparing represent 17% and 5% of the standardized care cycle costs, respectively.

Table 3. Fuzzy logic time-driven activity-based costing estimations of costs of the RA care cycle per care delivery value chain step

| | (a) Fuzzy logic cost estimates | | | (b) Moon El | (c) Non El | Total costs | % of |
|----------------------------|--------------------------------|-----------------------|----------------|--------------|--------------------------|-------------|-----------|
| | Smallest (€) | Most promising (€) | Largest (€) | estimate (€) | (c) NON-IL costsa (€) | [B+C] (€) | the total |
| Diagnosing | 93 | 143 | 217 | 151 | 331 | 482 | 32.2 |
| Preparing | 48 | 65 | 83 | 65 | 15 | 80 | 5.3 |
| Intervening | 91 | 148 | 189 | 143 | 187 | 329 | 22.0 |
| Recovering | 51 | 79 | 115 | 82 | 267 | 347 | 23.3 |
| Monitoring and managing | 42 | 63 | 88 | 64 | 192 | 256 | 17.1 |
| Total | 325 | 498 | 692 | 505 | 992 | 1497 | |

FL fuzzy logic

a) direct, b) indirect, c) honorarium direct

The costs related to both (medical) staff and other resources are shown in Fig. 2. Consultations with rheumatologists account for two-fifths of the total costs. In chronic care settings such as rheumatology, patients visit healthcare facilities often and therefore the contribution of consultations to the total costs is significant. In particular, this holds true for the first year of the RA standardized care cycle since patients have to visit the hospital frequently for consultations and medical tests. Almost one-third of the costs are spent on diagnostics and pharmacy, including drug costs. Over 60% of the CDVC recovering phase costs are due to pharmaceutical and diagnostic costs. Since patients generally switch to higher priced biologicals in case of ineffective treat-to-target after 6 months, pharmacy expenditures are relatively high. The physician assistant and nurse practitioner generate over 15% of the costs and the rheumatology nurse 8%. The care delivery costs spent on the counter and doctor's assistant account for less than 5% per cost driver.





FL-TDABC versus TDABC (Standard Times) and ABC

Based on the FL-TDABC approach, given the minimum, most promising and maximum time estimates, annual costs per RA patient range from a minimum of €1317 via a most promising value of €1490 to a maximum of €1684. This outcome reflects the real-life variation in patients' treatment regarding case-mix and multimorbidity, with a 22% difference between the minimum and maximum annual costs of RA treatment. The costs of the standardized RA care cycle totaled €1609 per patient based on TDABC and €1604 (SD €2139) per patient following the ABC methodology (i.e. DTC method). The variance with the average FL-TDABC costs of €1497 is explained by the difference in time commitment by the rheumatologist (consults) in the diagnosing and intervention

phases of the CDVC. FL-TDABC uses an average of 57 min consulting time by the rheumatologist, whereas the standard time is 130 min in those phases.

Statistical Analysis

The one-sample t test showed a significant difference in costs between the FL-TDABC and ABC (DTC) methodology at a 5% alpha level (p = 0.0101, t = 2.566). Thus, the findings of the one sample t test of FL-TDABC versus ABC, the cost accounting methodology currently applied in Dutch healthcare, suggests there is a significant difference in cost estimates. Concerning the difference between the TDABC and ABC (i.e. DTC) cost accounting methods, no significant difference was found on a 5% alpha level (p = 0.895, t = -0.131).

Discussion

Since TDABC estimates for care cycles are generally cumbersome to generate (through time studies) and do not allow for the variation in time spent on patients by healthcare providers, FL-TDABC is suggested as a viable alternative for cost allocation in healthcare. FL-TDABC highlights the time spent and costs related to the practice variation observed, through a relatively simple and quick querying of the relevant medical staff. The FL-TDABC model applied to the standardized RA care cycle at Maasstad Hospital shows that the diagnosing phase accounts for the bulk of the costs (32%), followed by the recovering and intervening phases (45.3% combined). Major cost drivers are consultations, more specifically with the rheumatologist, and pharmacy costs (i.e. medication). Maximum RA cycle costs for the more severe or multimorbid RA patients are 22% higher than the minimum costs, representing the less complex patients with shorter consultation times, observed using the FL-TDABC method.

TDABC based on standard times resulted in higher costs compared with the FL approach. Higher costs of the diagnosing phase in the TDABC model, largely explain the observed discrepancy between the models. As mentioned in the Methods section, ABC costs are derived from pricing DTCs. Total unadjusted TDABC cost estimates proved to be the highest, although the difference with the ABC (DTC) estimates are minimal. The FL-TDABC methodology allows for a more precise cost estimate, as the deterministic time estimates are pooled to an average and corrected for the subjectivity of variation in point estimations [21]. Furthermore, time estimates are obtained from healthcare providers who have first-hand knowledge of the patient population in terms of the required care. This leads to a better representation of the clinical daily practice that is incorporated in a value-based accounting method. Enhancing the TDABC methodology with FL allows for adjusting the subjectivity of time estimates [8]. Furthermore, the time-consuming process of observing activities and obtaining time estimates is avoided [8]. Time estimates per healthcare activity are based on the experience of multiple staff members, and through calculation of the fuzzy numbers a more objective cost estimation can be obtained. For the traditional ABC approach underlying the Dutch DTC system, the time spent and the cost price per practitioner are not disclosed. Therefore, the ABC approach is experienced as a black box, as DTC prices are not explicitly linked to the cost drivers of healthcare [22]. From an operational perspective, managing on the basis of activities is therefore more intuitive than on the DTC codes. Furthermore, the TFN numbers allow for a distinction between lowest, most promising (most frequent) and highest values, which can be linked to patients requiring low attention in terms of consultation time, as well as average- and high-demand patients. To conduct cost calculations for complex patients, the highest possible values will provide an initial estimate. Financial departments of hospitals can benefit from these insights. Moreover, the results of the current study can be used to examine a fair bundle payment rate for an episode of care. A third strength of the study is the broad range of costs (e.g. resources, activities and pharmaceutical) that are incorporated in the analysis, detailing the different factors associated with RA patient care. Furthermore, since the (standardized) patient journey is used as the basis for the calculation, activities performed by staff members not specifically taken into account in the DTC (i.e. ABC) methodology are also included.

A limitation of this study concerns the fact that the care cycle predominantly considered RA costs that are attributable to the hospital. However, the impact of chronic diseases on healthcare expenditures also reaches beyond the hospital silo and involves general practitioners and homecare. A second limitation is caused by the cost pricing system utilized in The Netherlands to reimburse healthcare expenses, as DTC codes do not include all healthcare activities and providers in relation to the healthcare provided, therefore the results of the ABC and (FL)-TDABC models are not fully comparable. This implies that the difference between the ABC and FL-TDABC annual costs could actually be higher than €186 per RA patient. In addition, as previously mentioned, the assumptions underlying the calculation of the DTC prices are not disclosed. Another limitation is related to the FL time estimates of the first consultation and the diagnosis consultation with the rheumatologists. Since it was unclear whether the time estimates between the ABC, TDABC and FL-TDABC estimates might be closer in proximity.

In comparison with the existing literature on this subject, a less time-consuming approach is suggested through the use of FL estimates. Staff members were interviewed, and, based on the pooled results, estimates are directly applied in the calculation, without expert panel groups or using the Delphi method to reach consensus [8]. The Delphi method, required to conduct traditional TDABC, involves other limitations concerning generalizability, validation and panelist satisfaction [23]. Additionally, the FL estimates were limited to the more complex activities within the care cycle, where practice variation between patients is most prominent as opposed to analyzing every activity such as a laboratory test.

Applying FL-TDABC has increased the precision and transparency in the costs of the RA care cycle and has provided insight into the cost drivers of the care delivery. Moreover, the model gave insight into the emphasis of the patient journey concerning the expenditures. Performing the (FL)-TDABC steps allows for an in-depth valuation of the practice variation regarding the activities and resources in care delivery, and therefore the model can be recommended as a cost accounting model in healthcare [8].

Potential reductions and optimization in terms of costs might be accomplished by shifting follow-up consultations from the relative costly rheumatologist to, for example, physician assistants and nurse practitioners. A previous study showed that patients are receptive to care delivery by physician assistants and nurse practitioners [24]. This applies, in particular, for the management and monitoring phase, since Dutch patients indicated that the lower-level complex care could potentially be provided by the physician's assistant [25].

With respect to the pharmacy expenses and in the light of minimization of excess healthcare delivery (i.e., waste reduction), close and remote monitoring of therapeutic drug levels may offer a personalized approach and address over- or undertreatment of RA patients [26]. As a result, efficiency gains affecting health outcomes and costs could be attained. Moreover, these results provide management and decision makers with information about the value in terms of cost efficiency related to the healthcare services patients require.

Conclusion

Defining healthcare costs through FL-TDABC is relevant from a VBHC perspective, since the time spent, and costs related to the practice variation observed, can be more quickly and adequately linked to the activities, including all relevant providers in the care cycle. Future research will focus on the long-term effects of RA expenditures, differentiating between single morbid and multimorbid patients. In addition, research in terms of a hybrid model considering a more complete allocation of costs to the medical staff within the current system will be investigated.

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Appendix

Supplementary file 1. Diagnosis-treatment combinations (DTCs)



3





Meso level: healthcare organization



Chapter 4

Financial Impact of Clinical Misregistration in Rheumatoid Arthritis Patients

Koster F, Kok MR, Kok MR, Conijn C, Weel-Koenders AEAM and Lopes Barreto D

Submitted
Abstract

Objective

Inaccurate registration of clinical diagnoses in electronic health records are common. Misclassification can adversely affect the treatment of patients as well as the research and benchmarking of health outcomes and costs. The objective of this study is to analyze the magnitude and the implications of misclassification within rheumatoid arthritis (RA) care from a clinical and financial perspective.

Methods

A retrospective cohort study was conducted to investigate the occurrence and effects of misclassification on the healthcare expenditures of RA patients within a Dutch hospital. RA patients were identified based on their classified treatment codes and verified through a patient by patient review of the associated electronic health records (2010-2017). Expenditures between the accurately classified patients and misclassified patients were compared within the first year of diagnosis and more than one year after the diagnosis. Additionally, regression analyses were performed to estimate the relationship between misclassification, expenditures and quality of life.

Results

Misclassification of RA diagnoses occurred in 18.8% of the patients. Over the seven year time period, the costs per patient were on average €6732,- higher for misclassified patients, predominantly caused by increased outpatient care delivery. In the year following the initial diagnosis, inaccurate registrations raised expenses of both inpatient and outpatient care and diagnostics compared with correctly classified patients. Over the seven year period, drug costs were higher for properly classified patients, but their improved health-related quality of life resulted in lower overall expenses.

Conclusions

Despite the fact that accurate registration of diagnoses in electronic health records is essential for patient-centered healthcare delivery, misclassification often occurs and it affects healthcare expenditures and the quality of patients' lives negatively. Therefore, more attention should be given to accurate registration of diagnoses, especially in chronic care delivery.

Introduction

With the increasing utilization of electronically recorded routine health data in hospitals, it has become essential to evaluate outcomes and costs in order to determine the added value of the delivered healthcare [1] [2] [3]. For effectiveness and efficiency purposes of healthcare interventions, the patient-reported health outcomes as well as the associated costs should be recorded accurately. Unfortunately, inaccurate registration of diagnostic codes in electronic health records (EHR) is fairly common [4]. EHR systems were primarily introduced to facilitate the correct recording of patients' medical information as well as the exchange of that information during the patient journey [5]. However, the reliability and value of EHR's is dependent on the available system codes, in addition to proper registration of the diagnosis by the care providers. From current literature it is evident that the typical limitations in interpreting causal relationships in cohort data stem mainly from the misclassification where it may also affect the interoperability of cost evaluations in hospitals. Data quality issues especially concern chronic diseases, such as Rheumatoid Arthritis (RA), in which the diagnosis can be a complicated process and is often made over time [6]. To support a holistic and integrated care delivery, where the accuracy of registration is depend on the available diagnostic codes and the treatment team, proper registration is essential [7]. Furthermore, adequate registration of the delivered healthcare, serves as a base for obtaining epidemiological frequency measures, enhancing transparency of the reimbursement and financing of healthcare expenditures and research purposes [8].

The prevalence of misclassification was previously studied at the outpatient rheumatology department of the Maasstad Hospital in the Netherlands, finding that some 25% of the patient files showed inaccuracies [9]. Misclassification of diagnoses was predominantly the result of wrong entries or miscoding in the EHR, the relationship between prompt treatment initiation and the time needed to properly diagnose a condition, or the incorrect conversion of diagnoses from the Dutch Diagnose Treatment Combination (DBC) into the International Statistical Classification of Diseases and Related Health Problems-version 10 (ICD-10) codes [9]. Misclassification regarding the health status and treatment of patients causes potential bias with respect to the benchmarking of (patient-reported) outcomes [10] [11]. However, to our knowledge, research on the economic impact of the misclassification concerning the RA diagnosis in The Netherlands is absent.

Understanding the economic implications of diagnostic misclassification in EHR's is especially relevant to gain better insight into the total costs of the RA care cycle, given

the growing number of patients suffering from rheumatoid diseases and the associated costs. From a value-based healthcare perspective, an accurate diagnosis promotes treating patients in a manner that is both timely and appropriate to their specific needs, which contributes to the overall goal of improving patient-centered care delivery and health outcomes. Moreover, to optimize the use of cost information in healthcare for e.g., cost-effectiveness analyses, misclassification should be diminished. Therefore, the objective of this research is to investigate the healthcare outcomes and costs associated with the misclassification of ICD-10 codes in RA patients in The Netherlands.

Materials and Methods

Study design and setting

For this retrospective cohort study we used data from the previous study at Maasstad Hospital concerning the reliability of ICD-10 codes for the registration of RA patients [29]. The data concerned the period 2010 through 2017 and encompassed all diagnoses of patients and the healthcare resources used. The EHR was implemented in the Maasstad Hospital in 2010. Data from 2018 onwards were not included due to the fact that a structural improvement cycle regarding the EHR (mis)classification was initiated with the medical staff. Ethical approval for this study was provided by Maasstad Hospital (JOINT Evaluation study, T2016-76).

Study population

Patients were identified by ICD-10 codes that correspond to a RA diagnosis e.g., codes M05, M06, M13.1, M13.9 or M13.0. ICD-10 codes M05 and M06 refer to RA, M13.1 matches the diagnosis mono-arthritis, M13.90 with oligo-arthritis and ICD-10 code M13.0 with poly-arthritis.

Data sources, collection and outcome measures

The data were retrieved from patients' EHR's and the financial records obtained from the finance and control department related to the provided treatment at the department of rheumatology and clinical immunology such as diagnostics, outpatient care and laboratory. Demographics and clinical data concerning age at diagnosis, gender, multimorbidity, disease duration, disease activity, medication use, and Euro Quality of Life – 5 Dimension (EQ-5D), a standardized measure of health-related quality of life, were collected as well. Disease activity was derived from the Disease Activity Score 28-joint count C-reactive protein (DAS28CRP) at time of the diagnosis [12]. The number of diagnoses in addition to the inflammatory arthritis diagnosis, was included and

defined as multimorbidity. Mean EQ-5D scores were calculated per patient ranging from 0 to 1, where higher values indicate a better quality of life, to gain insight in the health-related quality of life as perceived by the patients [13].

Primary outcome measure

In this study, clinical misclassification of diagnoses was defined as patients having a EHR classified RA diagnosis whilst suffering from other rheumatologic diseases. Analysis on the misclassification of the diagnoses was conducted in two different groups, the RA group (M05 and M06) and the other specified arthritis group (codes M13.1, M13.9 and M13.0) since misclassification occurs often in both of these disease populations. To assess and verify the number of misclassified cases within each group, the ICD-10 code was compared to the diagnosis as established by the rheumatologist (through per patient screening of the EHR). The clinical diagnosis as established by the rheumatologist was considered as the golden standard. Patients were assigned to the following groups: properly classified group consisting of the true positive group, i.e. RA according the rheumatologist and true negatives, patients who were not diagnosed with RA and with ICD-10 code that did not correspond to RA. Misclassified patients comprise the false positives which includes RA patients that according to a rheumatologist had no RA. And the misclassified patients with respect to the other specified arthritis group, false negatives, i.e. people suffering from RA who were not classified as RA patients. Experienced researchers assessed (NC and TK) and verified the EHR classified diagnoses and categorized patients into the misclassified or properly classified group (see supplementary files). Ten percent of the dataset was cross-checked and if ambiguity was present, a rheumatologist was consulted (AW and MK).

Secondary outcome measures

Expenditures were analyzed from a healthcare perspective, incorporating the medical costs of patients but excluding first-line care. The direct healthcare costs were based on the cost prices of the medical procedures. The components of the healthcare care utilization examined included emergency care, outpatient care, drugs, diagnostics, laboratory services, inpatient care and other medical services such as ergo therapy and physiotherapy. All healthcare expenditures were taken into account, incurred within the rheumatology department as well as costs from other departments within the hospital. Methotrexate - the first line therapy - costs, are however not included in the analysis as the dispenses of methotrexate were not properly registered in the EHR included in the EHR. Other drug costs, such as the costs of biologicals are taken into account in the analysis if these drugs were prescribed via the hospital pharmacy. Healthcare costs were calculated based on the overall medical services provided to the patients. Analyses were

stratified by group, and broken down in seven cost categories such as inpatient care, laboratory and outpatient care. The healthcare costs (euros) were indexed to the base year 2018.

Statistical analysis

Descriptive statistics are used to summarize the data. Continuous data are depicted as means and standard deviations or median and ranges depending on the distribution of the data. Categorical data are presented as percentages. The Wilcoxon rank sum test was performed if the data were not normally distributed, otherwise the t-test was used. All analyses were performed for the distinguished diagnostic classified groups. First, the medical resource usage was determined for the misclassified group, i.e. the false positives and false negatives, versus the properly classified groups. Resource utilization was examined within the first year of the initially classified ICD-10 diagnosis and more than one year after that diagnosis.

To examine the effect of the costs in the misclassification group and the correct classified group an Ordinary Least Squares (OLS) regression was conducted. Moreover, OLS regression analyses were performed to examine the effect of the health-related quality of life (EQ-5D) on the expenditures and adjusted for gender, age, multimorbidity and disease activity. If patients' EHR's had no information with respect to the medical cost, they were excluded from regression analyses when the type of missing data were confirmed to be at random. The number of observations per analysis are denoted in the tables. Data analyses were conducted using StateSE 15 for Windows, where a p-value of <0.05 was considered statistically significant.

Results

Patient characteristics

The study sample consisted of 802 newly diagnosed patients of which 70.8% is female (table 1). Overall, patients were on average 57.7 years old at the time of the RA diagnosis (SD=15.1). In the correctly classified group, the mean age was lower compared with the incorrect EHR diagnosis group (p<0.01). Multimorbidity, defined as the presence of >1 disease at baseline, was present in 75.6% of the patients. Frequently occurring multimorbidities were related to the specialisms of oncology, surgery, medical rehabilitation and gastroenterology. For both patient groups, the disease activity score indicated that the majority of the RA patients were in remission. Mean EQ-5D scores are highest in the properly classified group (mean=0.75). With respect to the other parameters, no

statistical significant differences were present. The frequency of misclassification was 18.8% with respect to RA patients. The number of true positives was 671 and 5 patients were true negatives. Concerning the false positives 106 patients were identified, against 20 false negatives.

| | | Properly classified patients | Misclassified patients | Total |
|---------------------------|------|------------------------------|------------------------|-------------|
| | | (n=676) | (n=126) | (n=802) |
| Gender female (n / %) | | 486 (71.9) | 82 (65.1) | 568 (70.8) |
| Age at diagnosis* (years) | | 57.2 (14.8) | 60.8 (16.5) | 57.7 (15.1) |
| Disease activity score | | 2.4 (1.1) | 2.4 (1.0) | 2.4 (0.99) |
| (range 0-9.4) | | | | |
| Multimorbidity (n /%) | | | | |
| N | one | 167 (24.7) | 27 (21.4) | 194 (24.2) |
| | 1-5 | 307 (45.4) | 51 (40.4) | 358 (44.6) |
| e | 5-10 | 121 (17.9) | 29 (23.0) | 150 (18.7) |
| | >10 | 81 (12.0) | 19 (15.1) | 100 (12.5) |
| EQ-5D score* | | 0.75 (0.20) | 0.67 (0.25) | 0.74 (0.21) |
| | | | | |

Table 1. Baseline characteristics of the RA population.

Note. Data are presented as mean and SD unless stated otherwise; * p <0.01 ; Number of patient with an available EQ5D scores was 156.

Cost analysis of RA misclassification

The breakdown of the healthcare costs highlighted the contribution of drug costs on the total costs in all the categories within the first year of diagnosis (figure 1). More specifically, in the misclassified group, outpatient care (54%) represents a higher share of the total costs compared with the proper classified groups (27%). Expenditures as a result of laboratory tests, diagnostics and drugs are relatively higher for the properly classified group. With respect to the drug costs (other than methotrexate), expenditures are more than four times as high in the accurately classified patients compared to the misclassified patients. The contribution of emergency costs and other costs (e.g. physiotherapy) were minor for both groups.





Analysis of the costs more than one year after the diagnosis shows significant shifts in the composition of the provided care (cost drivers) in comparison with the first year after the diagnosis (figure 2). Outpatient care continues to be utilized more by misclassified patients (26%) than by properly classified patients (15%). In both groups the share of drugs costs has increased, with respect to the misclassified patients, a steep increase from 4% to 20% is observed. Drugs continue to account for the largest cost component in properly classified patients, followed by outpatient care. The share of inpatient care has increased from 18% to 24% in the misclassified patients (versus 19% in properly classified patients).





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Regression analysis

The results of the OLS regression (adjusted $R^2 = 0.48$) corrected for age at diagnosis, disease activity and multi-morbidity showed that the healthcare costs in the misclassified patient group are on average €6,732 higher compared to the properly classified group over the 7 year period, however no significant difference was found between the groups (p=0.525). This corresponds to a difference of around 10 percent. Suffering from more than 6 multimorbidities, results in significantly higher expenditures (€33,720 per patient) in the seven year time period (p<0.001) and more than 10 multimorbidities results in an average of €49,708 in the seven year period (p<0.001). Disease activity also had an significant impact on the costs, enhancing the costs by €7,570 per patient on average. The sample consisted of only 50 responses due to the fact that disease activity was recorded in only a few cases, impacting the multivariate analysis.



Figure 3. Impact of misclassification on expenditures (>1 year after diagnosis).

Quality of life

As denoted in figure 4, a higher health-related quality of life (EQ-5D) is associated with lower expenditures (p=0.088). However this is not a statistically significant effect. An increase of one point on the EQ-5D scale, leads to a decrease of 21,205 euros per patient in the period 2010 to 2017. The association between health-related quality of life and expenses is not statistically significant for misclassified patients (p=0.460). However, the unadjusted model explains only 1% of the variation in spending.





Discussion

Accurate registration of diagnoses promotes holistic and integral patient-centered care delivery. Nevertheless, misclassification occurred in nearly 19% of the Rheumatoid Arthritis diagnoses at the hospital researched. Within one year following the initial diagnosis, the inaccurate registrations increased expenses of outpatient care compared with properly classified patients. After the first year, inpatient and outpatient care and diagnostics expenditures for misclassified patients continued to exceed the expenditures of the accurate classified patients. Over a seven year time period, the total costs per patient were on average €6732,- higher in the misclassified group. Higher self-rated quality of life scores by the properly classified patients was associated with significantly lower healthcare expenses.

Due to the fact that a treat-to-target strategy is initiated as quickly as possible, drug costs were more than four times higher in the properly classified group of RA patients. To monitor and timely adjust the (drug) treatment, laboratory tests are frequently performed which explains the higher expenses in the first year after diagnosis. Outpatient costs are relatively high in the misclassified patients, which is probably caused by the fact that patients more often visit the rheumatologist, as these patients experience other complaints as a result of the misclassified diagnosis. Although the healthcare costs for the misclassified patients are higher, no statistically significant difference was found between misclassified and properly classified patients. In both the misclassified and properly classified patients were highly influenced by multimorbidity and disease activity.

The share of accurate RA diagnoses in the Danish and Finnish national registries (around 80%) is in comparison with the results in our study [14][15]. In this study, apart from positive predictive values, i.e. number of true positives, the effect of diagnosis accuracy is assessed in terms of expenditures and health-related quality of life. Also, the study population contains real-world data of patients diagnosed from 2010 to 2017, so the data is considered to be a representative sample. Moreover, the health-related quality of life is examined by a generic patient-reported outcome measure, the EQ-5D. A last strength of the study is the validation of the diagnosis per patient by researchers, increasing the validity of the results.

A limitation of the study is the fact that methotrexate is not taken into account in the analysis. Since methotrexate is the preferred first-line treatment for RA and in general the entire patient population is administered methotrexate, the costs will be underestimated in the study population. However, as the costs of methotrexate are relatively low with probably minor per patient variances, the effect on the total expenditures is expected to be minor. A third limitation regards missing values of the ICD-10 codes. The number of missing values is relatively large concerning the regression analysis, leading to missing information with respect to expenses. Especially with respect to disease activity the number of missing's was relatively high. We expect that these data are missing at random due to the fact that the baseline characteristics of both groups do not significantly deviate from each other, except for age at diagnosis.

The financial consequences of EHR misclassification in the Maasstad hospital RA population are highlighted in this study. Inaccurate registration of diagnoses occurs frequently and it increases healthcare spending through predominantly inpatient care treatments. Properly classified patients also had relatively lower expenditures on outpatient care in comparison with misclassified patients. Improved quality of life is associated with lower expenses for accurately classified patients. Since an accurate registration promotes treating patients at the right place and time, more attention should be given to the correct registration of diagnoses. As a result of the findings in this study, further research should focus on investigating the accuracy and validity of RA diagnoses in the Netherlands. Other disease areas should also be aware of the amount of misregistration of diagnoses. Accurate classification and registration of real-life clinical data are essential for the delivery of value based healthcare, based on the evaluation of treatments, patient-reported outcomes, and expenditures.

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Chapter 5

Multimorbidity Status and Annual Healthcare Expenditures of Rheumatoid Arthritis Patients: a Dutch Hospital-Centered Versus Population-Based Comparison

Koster F, Bakx PLH, Kok MR, Lopes Barreto D and Weel-Koenders AEAM Rheumatol Int (2023). https://doi.org/10.1007/s00296-023-05282-w

Abstract

The prevalence of multimorbidity among rheumatoid arthritis (RA) patients is increasing and associated with worse outcomes. Therefore, management of multimorbid patients requires a multidisciplinary approach. However, healthcare systems consist of monodisciplinary subsystems, which limits collaboration across subsystems. To study the importance of a multidisciplinary, integrated approach, associations between expenditures and multimorbidity are assessed in real-life data. Retrospective data on RA patients from a Dutch single-hospital are analyzed and compared to the Dutch RA population data. The Elixhauser index is used to measure the multimorbidity prevalence. Regression analyses were conducted to derive the relationship between multimorbidity, healthcare costs and self-reported quality of life (e.g. EQ-5D). When analyzing the impact of multimorbidity within RA patients in context of a single-hospital context, multimorbidity is only partially captured: 13% prevalence versus 24% of the Dutch population. Multimorbidity is associated with higher care expenditures. Depending on the type of multimorbidity, expenditures are €43–€5821 higher in a single-hospital and from €2259–€9648 in population data. Finally, medication use associated with chronic diseases and self-reported aspects of well-being are associated with similar increases in healthcare expenditures as multimorbidity based on hospital care. Within RA, a single-hospital approach underestimates the association between multimorbidity and healthcare expenditures as 43% of healthcare utilization and expenditures are missed. To overcome a single-provider perspective in healthcare and efficiently coordinate multimorbid patients, besides providing holistic care, professionals also need to use data providing comprehensive pictures of patients.

Introduction

Multimorbidity is frequently present at the onset of rheumatoid arthritis (RA) and the prevalence increases from 38 to 56.5% after 10 years [1]. Multimorbidity requires a different approach concerning the care delivery process in patients with RA as the clinical outcomes and response on treatments might be delayed or poor [2,3,4]. Multimorbidity is defined as the coexistence of at least two chronic illnesses in one patient, implying that RA patients with at least one other chronic disease, are multimorbid [5]. Multimorbidity is associated with lower survival, quality of life and affects treatment and therefore requires extensive and ongoing care involving a multi-disciplinary team of providers is presumably required [2]. Moreover, the available knowledge predominantly comes from investigations on multimorbidity within solely primary care or hospital (out- and inpatient) data. As a consequence, insight regarding the impact of multimorbidity on the full spectrum of healthcare costs including primary- and mental care is lacking.

The current fragmented structure of the healthcare system with facilities and departments acting as independent providers of care, impedes the integrated delivery of care for multimorbid patients. Instead, these independent providers each focus on treating single illnesses [6]. This single-illness approach causes a lack of collaboration and coordination across care settings and healthcare providers and thus may lead to errors, increased expenditures and worse health outcomes [6]. Additionally, physicians are primarily conscious of the frequently occurring associated morbidities within their disease area. By contrast, value-based healthcare (VBHC) pushes for patient-centered integrated care delivery, i.e. a holistic multidisciplinary approach. At present, a generic outcome set for adults has been established as well as a Dutch guideline including a decision aid for healthcare providers [7]. However, these outcome sets have not yet been linked to healthcare costs.

Insight in health resource use and costs of RA patients with multimorbidity helps to highlight the proper scope for multimorbid disease management through integrated care delivery. Prior prospective cohort research showed multimorbidity in RA patients is associated with increased healthcare use, higher expenditures and reduced work-related productivity [6,7,8]. A Scottish study showed that the annual costs significantly differed in patients with only RA compared to patients with RA and a single comorbidity [9]. In addition to expenditures, multimorbidity also impacts the quality of life of patients suffering from RA. A higher number of multimorbidities is associated with declining Euro Quality of Lifel-5 Dimension (EQ-5D) scores [10]. The goal of VBHC is to compete on the value added from a patient perspective [11]. This evaluation of costs and (patient relevant) health outcomes requires reliable data. However, the data are often limited to a hospital setting, and hence valuable information regarding outcomes and costs is potentially overlooked. The aim of this research is to highlight the importance of this information gap by studying the association between multimorbidity and the healthcare spending among RA patients from an integrated perspective (e.g. primary care, secondary and mental care) versus a single hospital. In addition, the association between quality of life, the expenditures and multimorbidity is examined to give a comprehensive overview of the effects of a fragmented healthcare system.

Materials and methods

Study design and population

A retrospective cohort study design was applied to analyze the number of RA patients suffering from multimorbidity and the related healthcare expenditures. The data were obtained from a single-hospital data (Maasstad hospital) and a set of population-wide data sources (Dutch RA population). Patients who received RA care in the hospital and were at least 18 years old were included. Furthermore, inclusion was based specialist-diagnose code (0324)-101 from the Diagnosis Treatment Combinations system, which is linked to International Statistical Classification of Diseases and Related Health Problems-version 10 (ICD-10) codes M05 and M06 in the electronic health record. Diagnosis treatment combinations are a diagnostic-related group (DRG) type of system used for hospital care reimbursement [12].The final population dataset contains data from over 63,000 RA patients; RA patients who did not receive hospital care for RA in 2017, or for whom this care was not registered as such were not included. Over 2500 patients were incorporated in the single-hospital dataset.

Data sources and collection

Two datasets were analyzed, from a single-hospital and from the Dutch RA population. The Dutch RA population data were retrieved from Statistics Netherlands, including patients from all hospitals based on the specialist diagnosis code 0324-101. The data were linked at individual level to data on outpatient medication use covered by the mandatory public health insurance scheme, and comprised information on hospital diagnosis and procedures, annual health care spending for ten categories of medical care including primary care, tertiary care, mental care [13]. Finally, the data are linked at the individual level to information on the demographics age and gender from the mandatory Municipal Registry.

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Subsequently, the data were linked to the Public Health Monitor 2016 of the Community Health Services, Statistics Netherlands and the National Institute for Public Health and the Environment, which contains information on smoking, Body Mass Index (BMI), and aspects of quality of life: self-rated health and functional limitations for a sample of the population using validated scales [14]. The Health Monitor comprises of a composite questionnaire as part of a national evaluation of the health of the Dutch population and 3,421 RA patients answered this questionnaire [14].

Single-hospital data were retrieved from the outpatient rheumatology department at the Maasstad Hospital Rotterdam, a teaching hospital treating the largest population of RA patients in the Netherlands. The components of the services included all procedures provided at the Maasstad hospital, both within and outside the rheumatology department, applying DBC costs to calculate hospital expenditures. Furthermore, patients were asked to fill out the EQ-5D questionnaires every six months to assess the quality of life, however, a limited proportion of the RA patients participated in the questionnaires since the implementation of the questionnaires started in the course of the year 2017. Therefore, as a complement, the 'Patient Global' Visual Analogue Scale (PG-VAS), an element of the Disease Activity Score (DAS), is examined. Ethical approval is received through the JOINT Evaluation study (institutional code T2016-76).

Definition of rheumatoid arthritis and classification of multimorbidity

In the EHR the ICD-10 codes M05 and M06 are registered by the physician and transformed to the Dutch reimbursement code 101. The diagnostic code is utilized to analyze the costs in both the hospital and population dataset. Multimorbidity is often used interchangeably with comorbidity, the expressions can be distinguished by the fact that in the definition of multimorbidity a dominant disease is nonexistent [15]. In this study, patients are defined as multimorbid when suffering from RA and at least one other chronic disease as defined by the Elixhauser Comorbidity index [16, 17]. Comorbidity indices are generally applied to predict the mortality, hospitalization and functioning of patients by considering the level of comorbidity [18]. We chose to use to the multimorbidities discussed by the Elixhauser index because the Elixhauser is superior to the Charlson Comorbidity Index [19]. The Elixhauser index encompasses 30 different diseases in contrast to 19 morbidities concerning the Charlson Comorbidity Index, both defined by a variety of ICD-10 codes [17]. To increase the generalizability of the study to other disease areas, illness specific indicators such as the International Consortium for Health Outcomes Measurement (ICHOM) standard set for inflammatory arthritis, are therefore not analyzed.

Cost and health outcomes analysis

The results of the single-hospital analysis are compared with the Dutch RA population dataset to analyze the discrepancies when solely focusing on a hospital setting provided in one outpatient clinic. Further, the effect of multimorbidity and the quality of life of RA patients is evaluated, by the EQ-5D and self-reported measures of well-being (subsample of the Dutch RA population) [13, 22].

Statistical analysis

Descriptive statistics are used to summarize both the hospital and population data. To examine the effects of multimorbidity on the total costs of the healthcare provided to the patients, Ordinary Least Squares (OLS) regression were conducted for both datasets separately. The linear regression model examined the association between multimorbidity and the healthcare costs (dependent variable) by adjusting for age (categorical variable) and gender (binary variable). Disease duration was unavailable in the Statistics Netherlands dataset and is therefore lacking in the population analysis. An alpha level of 5% is considered with respect to the significance levels. To determine the type of missing data, missing cases are analyzed and reported. Data analyses were conducted using the statistical software packages R and StateSE 15 and 16.

Results

Descriptive statistics

In Table 1, the characteristics of the RA patient populations concerning the Maasstad hospital and the (Dutch) Statistics Netherlands data are presented. Over two-thirds of the patients in both samples is female. The overall Dutch population (64.3 years, SD=13.5) was on average older than patients in the single-hospital data (59.6, SD=14). In the single-hospital data, for less than 1% of patients, costing data were missing.

 Table 1. Descriptive statistics Rheumatoid Arthritis populations

| | Single-hospital data | Dutch RA Population |
|----------------------------|----------------------|---------------------|
| Number of patients | 2,582 | 63,851 |
| Female | 1,871 (72.4) | 44,320 (69.4) |
| Age, years (mean \pm SD) | 59.6 (14) | 64.2 (13.5) |
| <50 | 636 (24.7) | 9,082 (12.2) |
| 50-59 | 647 (25.1) | 12,528 (19.6) |
| 60-69 | 639 (24.8) | 17,781 (27.8) |
| 70-79 | 499 (19.3) | 16,501 (25.8) |
| 80+ | 159 (6.2) | 7,959 (12.5) |

Note. All data are presented as total number and percentages unless stated otherwise.

Multimorbidity prevalence

The overall percentage of patients suffering from multimorbidities based on the Elixhauser is 13.1% within the RA population of the single-hospital and 23.6% in the Dutch RA population data (Table 2). The distribution of the multimorbidities shows that cardiac arrhythmias and solid tumors (excluding metastasis) were the most common diseases in both populations, but the frequency is lower in the Maasstad hospital. In the Dutch RA population, the third most frequent morbidity was hypertension (uncomplicated), while obesity and uncomplicated diabetes for single-hospital were the third most frequent in the single-hospital data. Of all conditions in the Elixhauser Index, one-third did not occur at all in the single-hospital population.

| | Table 2. Percentage | patients s | uffering | from E | Elixhauser | Index | morbidities |
|--|---------------------|------------|----------|--------|------------|-------|-------------|
|--|---------------------|------------|----------|--------|------------|-------|-------------|

| Elixhauser Index components | Prevalence single-hospital | Prevalence Dutch RA |
|---------------------------------|----------------------------|---------------------|
| · | data | population |
| | (%) | (%) |
| Congestive heart failure | 0.8 | 2.2 |
| Cardiac arrhythmias | 2.9 | 6.1 |
| Valvular disease | 1.0 | 1.9 |
| Pulmonary circulation disorders | 0.2 | 0.6 |
| Peripheral vascular disorders | 0.1 | 1.7 |
| Hypertension, uncomplicated | 0.0 | 4.6 |
| Hypertension, complicated | 0.0 | 0.2 |
| Paralysis | 0.0 | 0.3 |
| Other, neurological disorders | 0.1 | 1.0 |
| Chronic pulmonary disease | 0.0 | 1.3 |
| Diabetes, uncomplicated | 1.6 | 3.0 |
| Diabetes, comp | 1.4 | 1.4 |
| Hypothyroidism | 0.6 | 0.7 |
| Renal failure | 1.3 | 1.6 |
| Liver disease | 0.3 | 0.6 |
| Peptic ulcer disease | 0.2 | 0.1 |
| Aids/HIV | 0.0 | 0.0 |
| Lymphoma | 0.5 | 0.4 |
| Metastatic cancer | 0.1 | 1.1 |
| Solid tumor, exc. metastasis | 1.7 | 5.7 |
| Coagulopathy | 0.2 | 0.3 |
| Obesity | 1.6 | 0.7 |
| Weight loss | 1.1 | 0.4 |
| Fluid and electrolyte disorders | 0.0 | 0.2 |
| Blood loss anemia | 0.0 | 0.2 |
| Deficiency anemia | 0.04 | 1.2 |
| Alcohol abuse | 0.04 | 0.0 |
| Drug abuse | 0.0 | 0.0 |
| Psychoses | 0.0 | 0.0 |
| Depression | 0.0 | 0.1 |
| Total | 13.1 | 23.6 |

Note. Based on Elixhauser index as described in Elixhauser et al. [36]

Cost analysis

Hospital expenditures over a 1-year period totaled ϵ 5417 per patient in the singlehospital data and ϵ 6419 in the Dutch RA population (Table 3). Hence, spending in the single-hospital is 87% of total hospital expenditures from the population perspective and 57% of total healthcare expenditures. Furthermore, the Dutch RA population data show that hospital care spending is approximately 67% of total medical care spending for this population (ϵ 9462).

When examining data on healthcare use from outside the hospital, as acquired from the Dutch RA data, the percentage patients receiving care that suggests multimorbidity is considerably higher (Table 3). Approximately half of the RA patients use medication related to cardiovascular diseases, 18% for lung diseases and approximately 15% regarding mental health issues. Furthermore, 3.5% of the Dutch RA population used

| | Single-hospital data | Dutch population data |
|--|----------------------|-----------------------|
| Number of patients | 2,582 | 63,851 |
| Outcome measures | | |
| Healthcare expenditures (mean \pm SD) | N/A | 9,462 (12,352) |
| Hospital care expenditures (mean \pm SD) | 5,417 (8,887) | 6,419 (8,977) |
| Multimorbidity (medication use) | | |
| Cardiovascular disease | N/A | 55.6% |
| Diabetes | N/A | 10.2% |
| Mental health problem | N/A | 14.7% |
| Asthma, bronchitis, COPD | N/A | 18.7% |
| Multimorbidity (mental healthcare use) | | |
| Basic mental healthcare | N/A | 1.2% |
| Specialist mental healthcare | N/A | 2.4% |
| Mental healthcare total | N/A | 3.5% |
| Total (unique)* | N/A | 69.3% |

Table 3. Expenditures and percentage RA patients using medication and mental healthcare (extramural care)

Note. *In within this category

mental health care, of which 2.4 percentage point used specialist, more complex mental healthcare. Taken together, almost 70% of the Dutch RA population uses medication or mental healthcare or hospital care that suggest multimorbidity.

Association expenditures and multimorbidity

The adjusted OLS regressions regarding the Elixhauser comorbidity index show that having multimorbidities explain (R²) 44% of the variation in expenditures in the singlehospital data and 18% in the Dutch RA population (Table 4). Multimorbidity is associated with a larger increase in healthcare costs and is significantly higher in the Dutch RA data (€2259–€9648) than in the single-hospital data (€43–€5821), which captures only hospital care expenditures and only a subset of multimorbidities. Expensive conditions include peptic ulcer, i.e. €5821 and €7683 in single-hospital and Dutch RA population data respectively, drug abuse (€14935), depression (€13754) and psychoses (€22053), where the latter three morbidities are exclusively encountered in the population data (i.e. in other hospitals). Females experience higher expenses while the effect of age is limited or even zero after controlling for morbidities. However, the disease duration is related to increased expenditures in the single-hospital data (€1121: 2–5 years; €3043: >5 years). The unadjusted results, i.e. without disease duration, of the regression in the single-hospital data demonstrated similar effects with respect to the significance and magnitudes in relation to the adjusted regression (see supplementary materials).

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Table 4. OLS regression Elixhauser variables: healthcare expenditures and multimorbidity in hospital vs. Dutch RA population data

| | Maasstad Hospital expenditures in euros: | | population data expenditures multimorbidity | population data: Hospital care expenditures in euros (all multimorbidity indicators): | | population data: Total health care expenditures in euros (all multimorbidity indicators): | |
|--------------------------|--|---------------|---|---|-------------|---|--|
| - | Coefficient | p-value | Coefficient | p-value | Coefficient | p-value | |
| Intercept | 3722 | 0.000 | 4977 | 0.000 | 4927 | 0.000 | |
| Age | | | | | | | |
| <50 years | ref | ref | ref | ref | ref | ref | |
| 50-59 years | 156 | 0.677 | -344 | 0.011 | -292 | 0.005 | |
| 60-69 vears | -15 | 0.968 | -279 | 0.038 | -510 | 0.000 | |
| 70-79 vears | -670 | 0.100 | -551 | 0.000 | -1435 | 0.000 | |
| 80+ years | -1432 | 0.019 | 195 | 0.303 | -3133 | 0.000 | |
| Gender (female) | 625 | 0.037 | 832 | 0.000 | 166 | 0.02 | |
| Disease duration | | | | | | | |
| <2 years | ref | ref | | | | | |
| 2-5 years | 1121 | 0.007 | | | | | |
| >5 years | 3043 | 0.000 | | | | | |
| Congestive heart failure | 113 | 0.000 | 5500 | 0.000 | 3360 | 0 | |
| Cardiac arrhythmias | 72 | 0.000 | 4333 | 0.000 | 3137 | 0 | |
| Valvular disease | 39 | 0.871 | 4108 | 0.000 | 3574 | 0 | |
| Pulmonary circulation | 558 | 0.000 | 11424 | 0.000 | 6172 | 0 | |
| disorders | | | | | | | |
| Peripheral vascular | 3410 | 0.148 | 8055 | 0.000 | 6248 | 0 | |
| disorders | | | | | | | |
| Hypertension, | N/A | N/A | 6663 | 0.000 | 4790 | 0 | |
| uncomplicated | | | | | | | |
| Hypertension, | N/A | N/A | -305 | 0.835 | 438 | 0.638 | |
| complicated | | | | | | | |
| Paralysis | N/A | N/A | 13516 | 0.000 | 7976 | 0 | |
| Other, neurological | 222 | 0.417 | 6193 | 0.000 | 2656 | 0 | |
| disorders | | | | | | | |
| Chronic pulmonary | N/A | N/A | 7513 | 0.000 | 4633 | 0 | |
| disease | | | | | | | |
| Diabetes, uncomplicated | 92 | 0.090 | 6664 | 0.000 | 3962 | 0 | |
| Diabetes, comp | 65 | 0.078 | 4585 | 0.000 | 2259 | 0 | |
| Hypothyroidism | 226 | 0.287 | 4652 | 0.000 | 3666 | 0 | |
| Renal failure | 157 | 0.000 | 6531 | 0.000 | 4569 | 0 | |
| Liver disease | 35 | 0.528 | 6992 | 0.000 | 5629 | 0 | |
| Peptic ulcer disease | 5821 | 0.000 | 10510 | 0.030 | /683 | 0.053 | |
| Aids/HIV | N/A | N/A | 12360 | 0.000 | 367 | 0.832 | |
| Lympnoma | 43 | 0.019 | 8461 | 0.000 | ///1 | 0 | |
| Metastatic cancer | 405 | 0.000 | 11/86 | 0.000 | 9648 | 0 | |
| Solid tumor, exc. | 124 | 0.000 | 4244 | 0.000 | 3627 | 0 | |
| Ceagulapathu | | 0.474 | 0714 | 0.000 | C A C A | 0 | |
| Coaguiopatny | 200 | 0.474 | 0714 | 0.000 | 0404 | 0 | |
| Weight loss | 209 | 0.000 | 5020 7100 | 0.000 | 5720 | 0 | |
| Fluid and electrolyte | -4 | 0.930 NI/A | 6687 | 0.000 | 4194 | 0 | |
| disorders | N/A | 11/74 | 0007 | 0.000 | 4157 | 0 | |
| Blood loss anemia | N/A | N/A | 5768 | 0.002 | 4160 | 0.001 | |
| Deficiency anemia | -284 | 0.580 | 5386 | 0.000 | 3521 | 0 | |
| Alcohol abuse | -1430 | 0.668 | -727 | 0.725 | -1140 | 0 483 | |
| Drug abuse | N/A | N/A | 14935 | 0.097 | 7023 | 0.082 | |
| Psychoses | N/A | N/A | 22053 | 0.002 | 3350 | 0.182 | |
| Depression | N/A | N/A | 13754 | 0.000 | 7221 | 0.016 | |
| Medication use | - | | | | | | |
| Cardiovascular disease | N/A | N/A | 1901 | 0.000 | 1118 | 0 | |
| Diabetes | N/A | N/A | -229 | 0.203 | -1004 | 0 | |
| Mental health problem | N/A | N/A | 2797 | 0.000 | 878 | 0 | |
| Asthma, bronchitis, | N/A | N/A | 1602 | 0.000 | 491 | 0 | |
| COPD | | | | | | | |
| ··· ·· ·· | | | 1 | | | | |

| Mental health care use Basic mental health care Specialist mental health care | N/A N/A N/A | N/A N/A N/A | 2311 6161 | 0.000 0.000 | 763 301 | 0.019 0.184 |
|--|-------------------|-------------------|--------------|----------------|------------|----------------|
| Ν | 2,552 | | 63,851 | | 63,851 | |
| F-VALUE | 74.91 | | 173.42 | | 103.43 | |
| P-VALUE | 0.000 | | 0.000 | | 0.000 | |
| R-squared (adjusted) | 0.44 (0.44) | | 0.2484 | | 0.183 | |

Note: N/A means no patients were registered/present within this category

Association quality of life, expenditures and multimorbidity

Multimorbidity as measured through healthcare utilization does not capture full health differences. Table 5 shows the results from separate regressions of total healthcare expenditures on a set of quality-of-life measures corrected for age and gender in the Dutch population data. The results show that these measures capture dimensions of well-being and health-related quality-of-life that are associated with much variation in healthcare spending in the Dutch RA population. A lower quality of life results in significant enhanced total healthcare expenditures within the patient population. For instance, a one-point higher BMI is associated with €191 higher health care expenditures. And the differences in healthcare spending that are associated with difference in self-rated health and functional limitations are even larger. The magnitude of the coefficients

| | | Share of the population | Coefficient | p-value |
|------------------------|------------------|-------------------------|-------------|---------|
| Self-rated health | Very good | 0.01 | (ref) | |
| | Good | 0.32 | -2412.5 | 0.184 |
| | Alright | 0.53 | 2313.0 | 0.200 |
| | Bad | 0.12 | 9184.2 | 0.000 |
| | Very bad | 0.01 | 13689.7 | 0.000 |
| Ν | | 3421 | | |
| Functional limitations | Severely limited | 0.15 | (ref) | |
| | Some limitations | 0.69 | -7897.7 | 0.000 |
| | Not limited | 0.16 | -11954.7 | 0.000 |
| Ν | | 3404 | | |
| Smoking | No | 0.85 | (ref) | |
| | Yes | 0.15 | 1231.1 | 0.042 |
| Ν | | 3334 | | |
| Ever smoked | No | 0.31 | (ref) | |
| | Yes | 0.53 | 1293.8 | 0.011 |
| | Yes, currently | 0.16 | 2139.2 | 0.002 |
| Ν | | 3185 | | |
| | | Mean (± SD) | | |
| BMI | | 26.12 (4.42) | 191.9 | 0.000 |

Table 5. OLS regressions of total healthcare spending quality of life measures in the Dutch RA population data

Note. Functional limitations are examined using the 7-item OECD limitations scale

reveals that the variation shown in this table is at least as large as the variation shown in the table with multimorbidity as measured by healthcare expenditures.

Patients in the single-hospital data filled out the EQ-5D questionnaire on quality of life measure, corrected for age and gender. Among the 58 patients in Maasstad hospital reporting an EQ-5D score in 2017, the mean score was 0.73 (SD \pm 0.19). The majority of the population scored between the 0.6 and 0.79 on the EQ-5D index, as identified by the researchers as an average to good quality of life. Poor quality of life, defined by the researchers as a EQ-5D score less than 0.40, is significantly associated with higher hospital expenditures while correcting for gender and age. The difference between experiencing an optimal quality of life (EQ-5D score equal to 1) and a health state equal to death (EQ-5D score equal to 0), in the single-hospital data lead to an average decrease of €14230 per patient (adjusted R^2 =0.14). In addition, the patients filled out the PG-VAS questionnaire (N=516): average score of 40.9 (SD±27.9) on a scale of 0–100 (adjusted R^2 =0.02). A higher PG-VAS score, i.e. more pain, resulted an increase of €24 per point (p=0.018), after correcting for age and gender.

Discussion

The total number of patients defined as multimorbid, i.e. suffering from RA and at least one additional illness, ranges from 23.1% (Dutch RA population) to 13.1% (single-hospital) when considering the Elixhauser index and up to 69% when measuring based on outpatient medication and types of health care expenditures that suggest multimorbidity. Expenditures from a single-hospital perspective make up 84% of the population hospital expenses and 57% of the total population. Multimorbidity is associated with higher healthcare expenditures, ranging from ϵ 43– ϵ 5821 in the single-hospital data and from ϵ 2259– ϵ 9648 in the Dutch RA population data.

A possible reason for the distinction in prevalence of multimorbidities is that RA patients in the Maasstad hospital (i.e. the single-hospital site) may be treated elsewhere in the region for diseases other than RA. Since the Elixhauser indices only include a subset of diseases and sources, the overall multimorbidity rate is expected to be underestimated in the study. This is confirmed by the data on outpatient medication use and health care expenditures that we analyze, which suggest that the prevalence of e.g. chronic heart disease, lung disease and mental health problems is much higher. In clinical practice, medical staff members should be aware of the possibility of missing information concerning multimorbidities. In RA patients suffering from multimorbidities, the

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adherence to the treat-to-target approach, which is recommended by the American College of Rheumatology and the European Alliance of Associations for Rheumatology, multimorbidities is often suboptimal [23]. As a result, treatment responses can be lower or delayed [23].

There are large differences in the costs between the single-hospital data and the Dutch RA population data. These differences are caused because patients may receive care in other hospitals and by other types of healthcare providers. For instance, healthcare, with the exception of some psychiatric care, is not provided at general hospitals in the Netherlands and therefore costs solely become visible when examining costs outside of the hospital. As patients suffering from chronic diseases such as RA have a higher risk to develop mental disorders such as depression, looking beyond the hospital division is recommended for providers of chronic patients [24, 25].

Apart from the differences in healthcare expenditures by multimorbidity as measured by diagnoses established when using healthcare, there is also variation in quality of life in the RA patient population. Like the use-based multimorbidity measures, this variation in quality of life is associated with variation in health care expenditures. Quality of life measures demonstrated worsened self-rated health, loneliness and functional limitations lead to increased healthcare costs. These findings are in line with previous research regarding health-related quality of life than the general population and that there is an inverse relation between quality of life and number and multimorbidities [26, 27, 28]. Although, different quality of life measures were applied in the data sources due to the availability of the instruments, the questionnaires utilized in the study are self-rated measurements and as shown in literature, measures depict parallel examinations of quality of life [29]. Therefore, the outcomes are suitable to compare.

Analyzing the impact of multimorbidity on the expenditures and health outcomes of patients in the context of a single-hospital, only partially captures multimorbidity. The results implicate that solely a hospital perspective may not be the proper scope for treatment, interventions and evaluations from a VBHC view. Furthermore, subjective measures such as self-reported quality of life offer a broader picture than multimorbidity measures based on healthcare use and are, like the use-based measures related to higher costs in multimorbid patients.

In comparison with the study of Gunderson et al. (2021) concerning the burden of multimorbidity in RA patients, our study included a significantly larger patient

population. Furthermore, the analysis performed in our study was not limited to examining the prevalence of multimorbidity within RA, but also examined the effects on costs and outcomes [1]. The incidence and prevalence of assessing multimorbidity was also based on inter alia the Elixhauser index [1]. In another study, the relationship between multimorbidity and healthcare costs in patients with musculoskeletal disorders, which also includes RA, was examined [9]. Besides impact on the direct costs as a result of hospitalization and hospital site visits, indirect costs (e.g. productivity losses) were also substantial within this patient population [9]. Although the researchers analyzed the association between healthcare costs, multimorbidity and quality of life, the focus was not specifically on RA patients,

Measuring costs from the whole spectrum of the care delivery process in the Netherlands is considered as a strength of this study. In a literature review conducted on mental health problems in patients with chronic illnesses, the authors also reported a positive association between multimorbidity and total costs [30]. The same conclusion can be drawn from a study performed in the United States, suggesting that RA suffering from depression are subject to higher healthcare utilization [31]. Similar advices and recommendations are found on the subject of multimorbidity and the proposed approach to move to a holistic practice of care delivery by incorporating elaborate data on expenditures and usage in different segments of healthcare [3, 28]. In this study, we demonstrated the effects on costs and utilization from the comprehensive perspective suggested. Additionally, addressing the challenge of multimorbidity by quantifying expenditures and the quality of life, is also viewed as an extension of the current literature, targeting for a complete picture of the multimorbidity inquiry instead of focusing on one element.

Limitations of the study include that for the Elixhauser index applied as multimorbidity proxy, a limited percentage of ICD-10 codes fall within the criteria set out in the index. Hence, the morbidities considered are not exhaustive for the whole spectrum of multimorbidity and might lead to an underestimation of the prevalence. In other studies, the reported prevalence of multimorbidity is therefore higher [1, 9]. A second limitation is that only a limited number of patients reported the EQ-5D score in 2017. The EQ-5D is an element of the Patient Reported Outcome Measures and the questionnaires have been implemented in 2017, explaining the low number of respondents. Another limitation is the fact that data from 2017 might not be generalizable to recent healthcare expenditures. On the other hand, the effect of multimorbidity on the expenditures and quality of life measures is not expected to extremely alter in a period of four years. Moreover, due to the variation in diagnosis registration with respect to different countries, the impact of

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multimorbidity on the expenditures will possibly vary per country [32]. For example, the prevalence of comorbidities in the United States in much larger than in the Netherlands. In addition, the geographic area and related socio-economic status is potentially of influence on the number of patients suffering from multimorbidities. As lower socio-economic is a predictor for the number of chronic illnesses prevalent in patients [33].

To optimize care delivery as part of the value driven care movement, insight in the variety of actors within a potential integrated practice unit (IPU) is relevant. In the end, this enables multidisciplinary steering on both the generic health outcomes and healthcare utilization (i.e. expenditures). The study addresses the knowledge gap concerning the impact of multimorbidity on the costs beyond the silos in healthcare. By analyzing their relevant multimorbidities and well-being, patients can be treated as separate entities instead of a cluster of single illnesses. A previous study examining the impact of multimorbidity on payment designs, demonstrated that healthcare reforms have to advance towards a coordinated care approach to deal with the current trends of growing multimorbid populations and to diminish the burden of patients who are coping with multimorbidity [34].

Furthermore, to provide an optimal and effective treatment to patients, physicians should be aware of the comprehensive picture of morbidities of patients besides the index disease. From an extramural perspective the uptake of medication showed multimorbidity impact on healthcare utilization. These additional sources on medication provide a more extensive insight in the impact of multimorbidity in RA patients.

To demonstrate the impact of the single-hospital focus, the goal of the study was to examine the costs and quality of life of multimorbid patients from a holistic point of view. This is achieved by comparing RA patient data from a Dutch single-hospital site with the national population data on the whole spectrum of care (i.e., primary, tertiary and mental care). Concerning the transition to a value-based reimbursement system, insight in the number of multimorbid patients from a national database is also relevant as for example in bundled payments, more complex patients have to be compensated for (e.g.by receiving an additional payment). As a result of the study, the question arises why multimorbidity is associated with higher costs; i.e. due to the differences in the RA treatment resources or underlying differences between patients. Future research will focus on defining the cause of the association.

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Appendix

Supplementary file A1. OLS regression Elixhauser variables: healthcare expenditures and multimorbidity in hospital vs. Dutch RA population data (unadjusted for disease duration in the single-hospital data)

| Maasstad Hospital multimorbidity indicators): multimorbidity Population data | Total health care expenditures: all multimorbidity indicators | |
|--|--|--|
| Coefficient p-value Coefficient p-value Coefficient | p-value | |
| Intercept 5540 0.000 4977 0.000 4927 | 0.000 | |
| Age | | |
| <50 years ref ref ref ref | ref | |
| 50-59 years 441 0.247 -344 0.011 -292 | 0.005 | |
| 60-69 years 527 0.170 -279 0.038 -510 | 0.000 | |
| 70-79 years -154 0.708 -551 0.000 -1435 | 0.000 | |
| 80+ years -894 0.082 195 0.303 -3133 | 0.000 | |
| Gender (male) -735 0.016 832 0.000 166 | 0.02 | |
| Congestive heart failure 116 0.000 5500 0.000 3360 | 0 | |
| Cardiac arrhythmias 73 0.000 4333 0.000 3137 | 0 | |
| Valvular disease 1 0.998 4108 0.000 3574 | 0 | |
| Pulmonary circulation 529 0.000 11424 0.000 6172 | 0 | |
| disorders | | |
| Peripheral vascular 3932 0.101 8055 0.000 6248 | 0 | |
| disorders | | |
| Hypertension, N/A N/A 6663 0.000 4790 | 0 | |
| uncomplicated | | |
| Hypertension, N/A N/A -305 0.835 438 | 0.638 | |
| complicated | | |
| Paralysis N/A N/A 13516 0.000 7976 | 0 | |
| Other, neurological 256 0.356 6193 0.000 2656 | 0 | |
| disorders | | |
| Chronic pulmonary N/A N/A 7513 0.000 4633 | 0 | |
| disease | | |
| Diabetes, uncomplicated 103 0.063 6664 0.000 3962 | 0 | |
| Diabetes, comp 55 0.143 4585 0.000 2259 | 0 | |
| Hypothyroidism 285 0.205 4652 0.000 3666 | 0 | |
| Renal failure 161 0.000 6531 0.000 4569 | 0 | |
| Liver disease 36 0.517 6992 0.000 5629 | 0 | |
| Peptic ulcer disease 5861 0.000 10510 0.030 7683 | 0.053 | |
| Aids/HIV N/A N/A 12360 0.000 367 | 0.832 | |
| Lymphoma 45 0.015 8461 0.000 7771 | 0 | |
| Metastatic cancer 386 0.000 11786 0.000 9648 | 0 | |
| Solid tumor, exc. 123 0.000 4244 0.000 3627 | 0 | |
| metastasis | | |
| Coagulopathy 53 0.491 8714 0.000 6464 | 0 | |
| Obesity 210 0.000 5026 0.000 3726 | 0 | |
| Weight loss -18 0.779 7199 0.000 4194 | 0 | |
| Fluid and electrolyte N/A N/A 6687 0.000 4197 | 0 | |
| disorders | | |
| Blood loss anemia N/A N/A 5768 0.002 4160 | 0.001 | |
| Deficiency anemia -173 0.740 5386 0.000 3521 | 0 | |
| Alcohol abuse -1034 0.0760 -727 0.725 -1140 | 0.483 | |
| Drug abuse N/A N/A 14935 0.097 7023 | 0.082 | |
| Psychoses N/A N/A 22053 0.002 3350 | 0.182 | |
| Depression N/A N/A 13754 0.000 7221 | 0.016 | |
| Medication use | | |
| Cardiovascular disease 1901 0.000 1118 | 0 | |
| Diabetes -229 0.203 -1004 | 0 | |
| Mental health problem 2797 0 000 878 | 0 | |
| Asthma, bronchitis, 1602 0.000 491 | 0 | |

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| COPD Mental health care use Basic mental health care Specialist mental health care | | 2311 6161 | 0.000 0.000 | 763 301 | 0.019 0.184 |
|--|-------------|--------------|----------------|------------|----------------|
| Ν | 2552 | 63,851 | | 63,851 | |
| F-VALUE | 74.68 | 173.42 | | 103.43 | |
| P-VALUE | 0.000 | 0.000 | | 0.000 | |
| R-squared (adjusted) | 0.43 (0.42) | 0.2484 | | 0.183 | |

Note: N/A means no patients were registered/present within this category

Acknowledgements

Results based on calculations by Erasmus University using non-public microdata from Statistics Netherlands. We thank the Community Health Services, Statistics Netherlands & National Institute for Public Health and the Environment for access to survey data from the 2016 Public Health Monitor (Gezondheidsmonitor 2016). The authors thank Hedwig Blommestein for comments on an early version of the paper.

Conflict of interest

None



Chapter 6

Evaluating the Implementation of a Patient-Reported Outcomes Dashboard: Effects on Health Expenditures and Outcomes

Koster F, Dikkenberg van den M, Kok MR, Lopes Barreto D, Weel-Koenders AEAM

Submitted
Abstract

Introduction

Dashboards depicting patient-reported outcomes (PROs) used in the consultation room are expected to enhance patient-centered care delivery. However, due to its intricate nature, evaluating these dashboards is challenging. To obtain insight into the added value of a PRO-integrated dashboard, the employment is assessed by evaluating outcomes and costs in patients with Rheumatoid Arthritis (RA).

Methods

The study population was derived from a retrospective cohort study concerning (N=173), using the JOINT decision dashboard displaying metrics such as PROs and clinician-reported outcomes. A control group, i.e., RA patients without supporting aid from the dashboard, was distinguished and comparative analyses were performed for PROs and expenditures. The study employed Generalized Least Squares (GLS) regression analyses to assess the impact of the dashboard. Analyses were adjusted for age, gender, multimorbidity and disease duration.

Results

The GLS regressions demonstrated RA patients in the dashboard group scored significantly better on the HAQ-DI (p=0.02) and EQ-5D (p=0.08) PRO's. PRO differences were also clinically relevant, as the minimally importance difference was reached. The dashboard group had on average \in 1133 lower healthcare expenses (p=0.073). From adjusted regressions, being female had a negative impact on the PRO scores (p<0.05) and multi-morbid patients had significantly higher expenses than patients without multi-morbidities (p<0.01).

Conclusion

Examining the effect of a dashboard stimulating the provision of patient-centered care from a broader perspective by means of a PRO dashboard, resulted in beneficial outcomes on PROs and expenditures. Future research should focus on qualitative measures regarding the content analysis of conversations held in consultation rooms.

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Introduction

Implementation of dashboards depicting Patient-Reported Outcome Measures (PROMs) in clinical practice and the increased number of campaigns related to shared decision making (SDM), have stimulated the dialogue between patients and providers regarding treatment options and preferences [1] [2]. Techniques developed to embed SDM within healthcare settings aim to share information regarding the relevant outcomes and care possibilities for a specific disease, given the physical clinical and patient reported condition and preferences of the patient [2]. By utilizing an outcome-based dashboard, healthcare providers can more easily integrate PRO measures into the patient journey for remote monitoring. Therefore, developing a dashboard that specifically addresses PRO implementation can be an effective strategy that adds value on both outcomes and healthcare cost, while enhancing patient-centered care at the right place [3].

Although PRO dashboards are increasingly being implemented in several disease areas, in daily practice, the uptake of both patient-reported and clinical outcomes in one dashboard is challenging, especially when the tools need to be seamlessly integrated without imposing additional time constraints on healthcare providers [4][5]. Moreover, significant hurdles such as the potential mismatch between the tool and specific clinical problems, and a lack of trust in the information presented, have also been identified as obstacles to their effective adoption [6][7]. As a result, the consequences of implementing PRO (decision) dashboards on for example healthcare utilization and outcomes are understudied [8][9].

As the number of patients with RA is expected to grow, the transition to patient-centered remote care delivery is of relevance as in the end, this might ease the capacity pressure on the healthcare providers. To our knowledge, until date, research is scarce with respect to the quantitative evaluation of dashboard utilization in the consultation room. To explore a broader spectrum of results that demonstrate successful collaborative deliberation, the aim of this study was to investigate the effects associated with the implementation of a patient-reported outcome dashboard in a chronic care setting by assessing both healthcare expenses and patient outcomes in Rheumatoid Arthritis (RA) patients [10].

Methods

PRO-based dashboard

To engage providers and patients in the personalized care process, in 2019 our decision dashboard was established and validated by the joint effort of patients and health care professionals from the Maasstad Hospital, Rotterdam the Netherlands and several IT partners (i.e. Microsoft Corporation and Macaw). The dashboard was considered by all stakeholders as a support tool to integrate the use of PROMs in the clinical practice, contributing to the process of patient-centered care since it allows for a broader insight into health status than sec clinician-reported outcomes such as laboratory results (supported by the Dutch National Health Institute, file number: 2018017132). The dashboard (called the Joint Decision Support), integrated in the electronic health record, displays the current and past information on rheumatology medication, relevant laboratory results, disease activity (DAS28CRP) as well as the results of the PROM questionnaires that patients fill out at least every six months [10]. Moreover, as a supplement to the PROs, four open-ended preparatory intake questions are appended to gain a precise insight the demands of the patient. The dashboard is fully integrated in the electronic health record and used during preparation of the consultation and the consultation itself. During consultations physicians turn their screen to the patient as a tool to support shared decision making.

Study design and population

This evaluation on the impact of outcomes and cost was performed in a retrospective reallife cohort of RA patients who were treated at the outpatient rheumatology and clinical immunology department of the Maasstad hospital, which is a top-clinical teaching and research hospital in Rotterdam, the Netherlands. In total, 9 rheumatologists, 1 nurse practitioner and 2 physician assistants participated. In February 2019 the PROintegrated dashboard was implemented. Data were retrieved in the period from August 2018 until December 2022. To examine the effect of the dashboard implementation, a control group and a dashboard (i.e., intervention) group were distinguished. The dashboard group consisted of patients that had used the dashboard in the consultation room and had filled out PROM before and during a follow-up PROMs at least 90 days after, but not more than 365 days after using the dashboard to filter the effect of the dashboard on the outcomes and costs properly (figure 1). The usual care group, i.e. control group, consisted of patients that filled out the PROMs in the half year (maximum of 180 days) before the implementation of the dashboard. Due to the nature of the inclusion criteria regarding the different time periods, it is possible patients are both present in the control and dashboard group.



Note. *180 before the implementation of the patient-reported dashboard. \P 90 to 365 days after implementation of patient-reported dashboard

Data collection

All data were collected during usual care as filling out PROMs was part of the standard care provision. As the study population consists of RA patients, the ICHOM standard set for Inflammatory Arthritis was utilized to select the relevant PROMs [9]. A total of five questionnaires are included in the dashboard: Functional Assessment of Chronic Illness Therapy (FACIT), Health Assessment Questionnaire-Disability Index (HAQ-DI), Rheumatoid Arthritis Impact of Disease (RAID), EQ-5D-5L and the Work Productivity and Activity Impairment (WPAI) for respectively the domains of fatigue, activity limitation, overall emotional and physical health impact and work ability and productivity [11]. Demographics were retrieved from the electronic health records, including age, gender, disease duration and multimorbidity. Age was defined as the age at the time of the consultation, disease duration is measured on January 1st 2022 and multimorbidity is defined as more than one diagnosis, besides RA as registered in the electronic health record. Health utilization encompassed outpatient- and inpatient visits, emergency room visits, laboratory tests, imaging and drug prescriptions concerning all departments at the Maasstad Hospital and were retrieved from the finance and control department. Costs were calculated based on the cost prices from the year 2019 of the retrieved procedures and care activities and were totaled the year following the actual face-to-face consultation.

Statistical analysis

Descriptive statistics for the variables gender, age, disease duration, multimorbidity status and Disease-modifying antirheumatic drug (DMARD) utilization were used to summarize the characteristics of the study population. Scores on the PROM questionnaires were examined as a proxy to assess the impact of the dashboard on 6

health-related outcomes. To assess the clinical relevance of the effects on the PROMs, the minimally important difference (MID) of these questionnaires was also evaluated. The MID is determined as the minimum change in a PRO score that impacts the outcome meaningful, either harmful or beneficial [12]. Previous research has shown that the MID for improvement of the HAQ-DI considering RA patients ranges from -0.09 to -0.24 [12][13]. Research on the evaluation of the MID estimate for EQ-5D amongst several illnesses including RA, corresponded in a score of 0.074 [14]. Generally, higher scores on the FACIT-F indicate lower quality of life, but in the hospital setting the outcomes were transformed into the opposite values. Furthermore, a Wilcoxon signed-rank test was performed to evaluate the statistical difference between the groups.

When groups are relatively small and costs tend to be positively skewed in general, the median is reported instead of the mean concerning the costs. Regarding the PROMs, scores also have a tendency to be skewed, and therefore medians are also reported. To analyze the effect of between the intervention group and the control group, a generalized least squares (GLS) regression was conducted to compare outcomes over time by exploring deviations in the healthcare intervention patient population and the control group. Due to the fact that a number of patients is included in both the control and post-dashboard group, a GLS regression model is applied. A GLS regression has the advantage that the model corrects for any correlation between the explanatory factors, contrary to the OLS method [14]. The GLS model is preferred over a generalized estimating equation (GEE) model since the sample size is relatively low. A GLS regression with a gamma distribution was also considered for the expenditures, however since the coefficients in GLS regressions with a gamma distribution are difficult to interpret, a normal distribution was applied. In total, five regression models were estimated, four concerning the effects on the health outcomes and another one regarding the expenditures. The outcomes and associated costs are subjected to distinct GLS analyses to ascertain the exact influence of the dashboard on these parameters.

Furthermore, adjusted regression analysis, accounting for the factors age, disease duration, gender and multimorbidity, were performed. StataSE (version 15) was applied to perform the statistical analyses in the study where a p-value of <0.05 is considered as statistically significant.

Results

Study sample

Table 1 displays the characteristics per group. Females are more represented in the control group (73.8%) compared to the dashboard group (62.0%). With respect to the mean age and disease duration, minor differences between the groups are observed. In the control group a higher percentage is suffering from multi-morbidities compared with the dashboard group (62.3% versus 55.5%). From the 61 patients in the control group, 25 patients are also included in the dashboard group (duplicate group). The characteristics from the duplicate patient group are similar to the control group population; on average the age is higher and the disease duration as well. DMARD utilization, including the use of biologicals was elevated in the dashboard group compared to the control group.

Table 1. Baseline patient characteristics

| | Control group | Dashboard group | Duplicate group |
|-----------------------------|---------------|-----------------|-----------------|
| | (N=61) | (N=137) | (N=25) |
| Gender (% female) | 73.8% | 62.0% | 76.0% |
| Age (mean, SD) | 60.6 (13.2) | 60.8 (11.9) | 62.6 (13.0) |
| Disease duration (mean, SD) | 11.1 (4.2) | 10.9 (4.2) | 11.3 (4.4) |
| Multi-morbid¶ (%) | 62.3% | 55.5% | 60.0% |
| DMARD utilization (%) | 70.5% | 78.8% | 76.0% |

Note. I more than one diagnosis

Patient-reported outcome measures

When comparing the average PROM scores of the control and dashboard group, the health outcomes on the EQ-5D, HAQ-DI and RAID are worse in the control group. The FACIT questionnaire however, showed that the control group scored worse in terms of fatigue compared to the dashboard group. Besides the statistical significant effect of found concerning the effects on the PROMs (HAQ-DI and EQ-5D), the MID of these questionnaires was also reached. The results of this study indicate an average decrease of 0.26 points per patient, suggesting a clinically relevant effect on the HAQ-DI score. The Wilcoxon Signed-Rank Test showed that the values of the EQ-5D, HAQ-DI and RAID statistically differed in the control group compared to the dashboard group.

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| Scores on questionnaires | Control group | Ν | Dashboard group | N | p-value |
|--------------------------|--------------------|----|--------------------|-----|---------|
| (median, IQR) | | | | | - |
| EQ-5D | 0.72 (0.47 – 0.81) | 46 | 0.79 (0.66 – 0.87) | 128 | 0.03 |
| FACIT | 20 (10 - 31) | 51 | 15 (7 – 28.2) | 113 | 0.11 |
| HAQ-DI* | 0.88 (0.38 – 1.33) | 50 | 0.63 (0.13 – 1.26) | 127 | 0.01 |
| RAID* | 4.6 (1.76 – 6.16) | 49 | 3.5 (1.39 – 5.78) | 122 | 0.02 |
| | | | | | |

Table 2. Patient-reported outcome measures

Note. *Higher scores indicate a lower quality of life

Healthcare utilization

Average costs are ϵ 7733 (SD: ϵ 10604) per patient in the control group, median costs per patient are ϵ 4745 (IQR: ϵ 1659 – ϵ 7712) per year. In the dashboard group, median yearly costs are lower, namely ϵ 2657 (IQR: ϵ 959 – ϵ 6999), the same holds for the mean costs, ϵ 6862 (SD: ϵ 12784). The majority of procedures, as measured by expenses, are performed at the rheumatology department; 36.8% in the post-dashboard group and 42.0% in the control group. In both groups, besides rheumatology, a large share of the expenses is related to orthopedic care delivery, especially in the control group. Noteworthy is the fact that expenses regarding pulmonary diseases are 10.8% in the post dashboard group compared to 4.1% in the control group. The number of provided care services in the dashboard group is on average 40 per patient compared with 60 in the control group (note: one laboratory test is equivalent to one activity). Mainly the activities that belong to the care path of a RA patient are more frequently observed in the control group, such as consultations, blood tests and injections for biologicals. On average patients in the dashboard group had 3.6 consultations at the rheumatology department (either electronic or physical) and in the control group the number of consultations was 5.7. The mean cost price of a consultation totaled ϵ 218.

| | Control group (N=61) | Dashboard group (N=137) |
|--|-----------------------|-------------------------|
| Total costs (SD) | €471,693 | €940,026 |
| Cardiology | €42,089 (€10,114) | €58,883 (€12,139) |
| Long | €19,407 (€3,712) | €101,404 (€19,697) |
| Orthopedics | €92,326 (€22,780) | €161,400 (€31,072) |
| Surgery | €17,792 (€5,187) | €44,874 (€26,269) |
| Rheumatology | €197,892 (€20,082) | €345,978 (€21,228) |
| Other departments | €102,187 (€8,268) | €227,487 (€14,427) |
| Mean costs per patient (SD) | €7733 (€10,604) | €6862 (€12,784) |
| Median costs per patient (IQR) | €4745 (€1659 – €7712) | €2657 (€959 – €6999) |
| Number of care services per patient | 60 | 40 |
| Number of consultations (rheumatology) per patient | 5.7 | 3.6 |

Table 3. Healthcare expenses in the Maasstad hospital

Note. N: number of patients SD: standard deviation, IQR: interquartile range

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Generalized least squares regression analysis

The results of the unadjusted regression on both the expenditures and the PROMs are displayed in table 4; model 1. Utilizing the dashboard in the consultation room has a positive effect on expenditures (p=0.073), on average patients in the dashboard group, have \in 1133 (~14%) lower healthcare expenses compared to the control group. Concerning the PROMs, the HAQ-DI showed a statistically significant effect on perceiving better health outcomes (– 0.26) in the dashboard group (p=0.021). Considering an alpha of 10%, the EQ-5D likewise resulted in an average increase of 0.09 points in the dashboard group (p=0.082). The FACIT and RAID questionnaires showed no significant impact on the outcomes when comparing the control and dashboard group. The results of the RAID questionnaire indicated similar outcomes, i.e., the dashboard group was more favorable in terms of outcomes, however, the effect was not statistically significant (p=0.154). Moreover, the FACIT corresponded with a decline in the score, suggesting a higher degree of fatigue in the dashboard group (p=0.115).

The results of the adjusted GLS regression analysis are also depicted in table 4, model 2. From the adjusted regression analysis, the conclusion can be drawn that multimorbidity significantly affects expenditures (p=0.005). The variables age, gender, disease duration and having support from the patient-reported outcome dashboard showed no significant effects on the health expenses. In GLS regressions on the health outcomes, being female had a significant negative effect on the quality of life according to the EQ-5D and RAID PROMs (p<0.01), however, a positive effect on the outcomes of the FACIT (p<0.001) and HAQ-DI (p<0.05) questionnaires. Moreover, aging had a slightly negative impact on EQ-5D score.

| | Model 1: Coefficient | 95%-CI | p-value | Model 2: Coefficient | 95%-CI | p-value |
|------------------|----------------------|----------------|---------|----------------------|-----------------|---------|
| | (SE) | | | (SE) | | |
| Costs | | | | | | |
| Intercept | €8250 (€1056) | | | €1546 (€5990) | | |
| Dashboard | - €1133 (€632) | €6180 - €10320 | 0.073 | - €640 (€807) | - €2222 – €942 | 0.428 |
| Female | - | | | - €96 (€2248) | - €4502 - €4310 | 0.966 |
| Age | _ | | | - €14 (€90) | - €190 - €163 | 0.876 |
| Disease duration | _ | | | €422 (€258) | - €83 - €927 | 0.102 |
| Multi-morbid | - | | | €4066 (€1437) | €1243 - €6848 | 0.005 |
| EQ-5D | | | | | | |
| Intercept | 0.62 (0.05) | | | 1.13 (0.13) | | |
| Dashboard Female | 0.09 (0.04) | - 0.01 - 0.18 | 0.082 | -0.02 (0.01) | -0.04 - 0.01 | 0.136 |
| Age | - | | | -0.14 (0.05) | -0.240.04 | 0.006 |
| Disease duration | - | | | -0.004 (0.002) | -0.010.00 | 0.039 |
| Multi-morbid | - | | | -0.00 (0.01) | -0.02 - 0.01 | 0.570 |
| | - | | | -0.03 (0.03) | -0.08 - 0.02 | 0.263 |
| FACIT | | | | | | |
| Intercept | 21.2 (1.76) | | | 13.2 (3.05) | | |
| Dashboard Female | - 3.34 (2.12) | -7.50 - 0.81 | 0.115 | 0.00 (0.00) | -0.00 - 0.00 | 0.999 |
| Age | - | | | 7.50 (2.13) | 3.33 - 11.68 | 0.000 |
| Disease duration | - | | | 0.00 (0.00) | -0.00 - 0.00 | 0.999 |
| Multi-morbid | - | | | 0.01 (0.25) | -0.47 - 0.50 | 0.959 |
| | - | | | 0.00 (0.00) | -0.00 - 0.00 | 1.000 |
| HAQ-DI | | | | | | |
| Intercept | 0.996 (0.09) | | | 0.12 (0.35) | | |
| Dashboard Female | - 0.26 (0.11) | -0.480.04 | 0.021 | -0.07 (0.05) | -0.17 - 0.04 | 0.213 |
| Age | _ | | | 0.34 (0.13) | -0.17 - 0.04 | 0.010 |
| Disease duration | _ | | | 0.01 (0.01) | -0.01 - 0.02 | 0.313 |
| Multi-morbid | - | | | 0.01 (0.02) | -0.02 - 0.04 | 0.566 |
| | - | | | 0.10 (0.10) | -0.08 - 0.29 | 0.276 |
| RAID | | | | | | |
| Intercept | 4.22 (0.42) | | | 2.25 (0.59) | | |
| Dashboard Female | - 0.60 (0.36) | -1.44 - 0.23 | 0.154 | 0.00 (0.00) | -0.005 - 0.00 | 1.000 |
| Age | - | | | 1.32 (0.41) | 0.52 - 2.13 | 0.001 |
| Disease duration | _ | | | -0.00 (0.00) | -0.00 - 0.00 | 1.000 |
| Multi-morbid | _ | | | 0.03 (0.05) | -0.06 - 0.13 | 0.479 |
| | _ | | | 0.00 (0.00) | -0.00 - 0.00 | 1.000 |

Table 4. Results of the generalized least squares regression analysis (model 1: unadjusted results,model 2: adjusted results)

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Discussion

The added value of a patient-reported outcome dashboard, depicting a variety of patient-reported and clinical outcomes in the consultation room, shows positive effects on both the healthcare expenditures and the relevant outcome on physical functioning (HAQ-DI) and health related quality of life (EQ-5D) in Dutch RA patients with a long disease duration. In-hospital healthcare costs were in patients using the JDS dashboard on average 14% lower compared to non JDS users. Additionally, patients in the JDS-dashboard group had more favorable health outcomes as demonstrated by a decrease in HAQ-DI scores and an increase in mean EQ-5D scores when compared to the control group. These results show the added value of the use of a patient-reported and clinical outcome dashboard in clinical practice improve patient outcomes while also reducing healthcare costs. The EQ-5D increase is perceived as clinical relevant.

The RAID and FACIT PROs suggested no significant difference between the control and dashboard group. However this can also be the result of face validation, indicating that it seems that the tests on the surface measure what the test should measure but in reality this is hard to verify. For the FACIT, this is likely caused by the fact that the FACIT focuses on a specific domain of the health-related quality of life, namely fatigue. Treatment of fatigue in RA patients is a complex and challenging symptom to treat in (RA) patients [15]. Research has shown that the impact on fatigue is limited as measured in RCTs is, which complicates to establish meaningful MID estimates for patients [15]. On the other hand, the dashboard group showed improvement in disease impact as expressed by RAID scores, though the difference was not statistically. For the interpretation, it is important to note that RAID scores are predominantly useful in discriminating patients with moderate to high levels of disease activity [16]. In this study, disease activity could not be taken into account as a result of the number of outdated information on disease activity in the patients. These findings indicate that the patient-reported outcome dashboard may have a greater impact on certain aspects of RA management than others, and future research could explore these nuances further.

Apart from the enhanced health effects illustrated, the hospital related expenses in the dashboard group were approximately 14% lower as well. The difference in expenditures can, for one-third of the total, be attributed to the fact that the patients in the control group had on average two more consultations at the rheumatology department. Furthermore, multi-morbidity is more prevalent in the control group, hence if patients are treated for these multi-morbidities in the Maasstad hospital, this could result in higher expenses.

This is also supported by the findings of the adjusted regression analysis, showing that multi-morbidity has a significant effect on the healthcare expenditures. Comparison with other studies is complicated as similar studies are not conducted until date.

As mentioned above, only limited number of studies has evaluated a patient-reported and clinical outcome dashboard in a quantitative manner. The emphasis of previous research in the field of RA is the practical implication of dashboard development and its use in the clinical practice [17][18]. A recent systematic review conducted by Dorr et al. (2022) demonstrated that only a limited number of the five studies have reported quantitative results on PROMs in non-rheumatology disease areas, with four studies showing no significant effects on outcome improvements in terms of PROMs and one study a minimal effect [19]. The present study takes a next step by evaluating the dashboard's impact on health outcomes and expenditures for patients beyond the narrow time perspective as outcomes were assessed in the year following the patientreported outcome dashboard utilization. An important step in the research regarding patient-reported outcome dashboard as the majority of the research is focused on short-term effects.

This study has notable strengths, including the statistical methodology chosen to evaluate the dashboard. Although a number of patients was represented in both control and dashboard group, a GLS regression model allowed for the correction of this bias [14]. Hence, a reliable indication of the implications of the dashboard on effects and expenditures could be given. Another strength is the application of PROMs to measure health outcome improvement in patients. This is especially important from a valuebased healthcare perspective, moving beyond a clinical view based on predominantly clinical parameters such as disease activity. Additionally these results can enhance the movement to remote monitoring of chronic diseases.

However, there are some limitations to the study that should be acknowledged. Limitations include the questions around the generalizability of the results given the small size of the study sample. Another appropriate research design to evaluate this intervention, is for example a differences-in-difference (DiD) model. DiD models aim to compare outcomes over time by exploring deviations in the healthcare intervention patient population and the control group, i.e. receiving no intervention. DiD models test statistical inference in the case when RCTs are infeasible [20]. However, in order to apply a DiD, a large study sample is required. A case control study was also not possible due to the fact a number of patients were included in both the control and dashboard group. A last limitation is related to the adjusted GLS regressions, the scores on the disease activity measures were outdated in the majority of the patients and could therefore not be incorporated in the GLS regression analyses.

In summary, the implementation of a patient-reported and clinical outcome dashboard in the consultation room of providers and RA patients, demonstrated positive effects on the EQ-5D and the HAQ-DI scores of patients. Furthermore, healthcare expenditures were lower when the dashboard was being used. The improvement in PROMs observed with the implementation of the dashboard, may induce a change in the patient's management' of RA. For instance, as result of an observed elevated score on the PROs, the tapering of medication in patients can be initiated. Future research is needed to evaluate the effectiveness of such dashboards in other (chronic) disease areas, shared decision making and in larger populations. 6

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Macro level: health policy

Chapter 7

Capturing Patient Value in an Economic Evaluation

Koster F, Kok MR, Lopes Barreto D and Weel-Koenders AEAM

Arthritis Care & Research (2023), https://doi.org/10.1002/acr.25229

Abstract

Objective

Economic evaluations predominantly use generic outcomes, such as Euro Quality of Life-5 Dimension (EQ-5D) to assess the health status. However, because of the generic nature, they are less suitable to capture the quality of life of patients with specific conditions. Given the transition to patient-centered (remote) care delivery, this study aims to evaluate the possibility to use disease-specific measures in a cost-effectiveness analysis (CEA).

Methods

A real-life cohort from Maasstad Hospital (2020-2021) in the Netherlands, with 772 Rheumatoid Arthritis (RA) patients, was used to assess the cost-effectiveness of electronic consultations (e-consultations) compared with face-to-face consultations. The Incremental Cost-Effectiveness Ratio (ICER) based on the generic EQ-5D was compared with ICER's based on RA specific measures; Rheumatoid Arthritis Impact of Disease (RAID) and Health Assessment Questionnaire-Disability Index (HAQ-DI). To compare the cost-effectiveness of these different measures, HAQ-DI and RAID were expressed in QALYs via estimated conversion equations.

Conclusions

The conventional ICER (e.g. EQ-5D) indicates that e-consultations are cost-effective with cost savings of - €161k per QALY gained for a prevalent RA cohort treated in a secondary trainee hospital. RA specific measures show similar results, with ICER's of - €163k per HAQ-DI(QALY) and - €223k per RAID(QALY) gained. RA specific measures capture patient-relevant domains and offer the opportunity to improve the assessment and treatment of the disease impact.

Discussion

Disease-specific patient-reported outcome measures (PROMs) offer a promising alternative for traditional measures in economic evaluations, capturing patient-relevant domains more comprehensively. As PROMs are increasingly applied in clinical practice, the next step entails modelling of a RA patient-wide conversion equation to implement PROMs in economic evaluations.

Introduction

Because of the adoption of technological innovations, conventional healthcare provision is shifting from hospitals and other facilities to the day-to-day life of patients. Results of earlier studies have shown the (cost) efficacy of e-health and telemonitoring solutions [1] [2] [3]. Patients with chronic conditions visit the hospital for check-ups regularly; Rheumatoid Arthritis (RA) sufferers approximately every three to four months. RA patients experience challenges in managing check-up appointments, transportation to outpatient clinics and their employability, due to relapses of the disease [4]. Although accelerated by COVID-19, the adoption of e-health and telemonitoring is perceived as a structural component of a broader transformation to patient-centered healthcare [1]. E-health and telemonitoring are considered to be promising methods to monitor RA patients remotely, thereby easing the burden for patients and promoting a shift toward a more patient-centered healthcare system [1] [5] [6]. A recent study by Bos et al. (2021) demonstrated that patients with rheumatic and musculoskeletal diseases (RMDs) were satisfied with telemedicine, i.e. telephone and video consultations, provided in the Netherlands [1]. However, quantitative results on patient-reported outcomes are lacking and therefore the added value in terms of healthcare effects and costs is still unclear.

In conventional economic evaluations within rheumatology and other diseases, the focus with respect to the assessment of the health-related quality of life as experienced by patients is predominantly on generic measures, such as the Euro Quality of Life- 5 Dimension (EQ-5D) or the Short Form Health Survey 36-Item (SF-36). However, utilities such as Quality Adjusted Life Years (QALYs), derived from the EQ-5D and SF-36, limit the patient perspective on health outcomes in economic evaluations as they are calculated on a more generic population basis and not specific to one health condition [7]. With the desired transition to more personalized and patient-centered care delivery, these instruments are less suitable to capture the full spectrum of patient relevant domains, as covered by for example the International Consortium of Health Outcome Measures (ICHOM) sets [8]. Patient relevant outcome domains are the cornerstone of the transition to a more value-based healthcare (VBHC) system, as embraced in several countries including the Netherlands [9] [10].

Although the EQ-5D is the preferred tool in cost-utility analysis, evidence concerning the applicability of the questionnaire in non-commercial, clinical practice is lacking [11]. The usability of the EQ-5D in clinical practice is limited due to the so-called ceiling effect, i.e. health scores clustering at the positive end of the scale [12]. Moreover, the EQ-5D was initially developed as a supplemental tool, in addition to more disease-specific patient-reported outcome measures (PROMs) [12]. As a result, the EQ-5D is used less often in clinical daily practice, which might influence the economic evaluation purposes of this questionnaire. However, patient relevant outcomes can be applied to broaden the current elements of value, e.g. QALYs and net costs, in health technology assessment (HTA) [13].

To incorporate the full spectrum of patient relevant domains as captured through PROMs and the transition to patient-centered (remote) care delivery in an economic evaluation, this study examines the impact on costs and perceived healthcare effects by RA patients engaging in electronic consultations (e-consultations). For this purpose, a comparison is made between the conventional economic methodology of valuing health by assessing the QALYs as obtained via the traditional measure EQ-5D versus the RA specific measures Rheumatoid Arthritis Impact of Disease (RAID) and Health Assessment Questionnaire-Disability Index (HAQ-DI).

Materials and methods

Study design and population

The real-life retrospective cohort of over 3,000 patients receiving RA care in the first quarter of 2020 (with a one-year follow-up) at the rheumatology department of the Maasstad Hospital in Rotterdam, the Netherlands, served as the base population for this study. Patients aged 18 years and over were included if RA was diagnosed by a rheumatologist at least one year earlier, and the patient's PROM and disease activity were available within 6 to 12 months after the consultation date [14]. Patients were excluded from the study if their PROMs were unavailable and/or their disease activity score (DAS) was lacking. This resulted in a study population of 772 patients (see supplementary file 1). As part of the study protocol (T2016-76) institutional permission is given to evaluate retrospective data.

Real-life (intervention) groups

The study population was split into two groups: RA patients using face-to-face consultations (control group) versus those with e-consultations (intervention group). E-consultations were defined as consultations performed via telephone and/or internet, the latter through a software service ("BeterDichtbij©"), made available by Maasstad Hospital as a secured (web-based) application [15]. Face-to-face consultations were defined as the patient visiting the healthcare provider at the hospital site. To minimize the difference in patients concerning their underlying disease characteristics (e.g.

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disease activity) between the two groups, the first consultation in the year 2020 was selected as the baseline measure point. If only one type of consultation, i.e., either an e-consultation or face-to-face consultation, occurred in 2020, the patient was assigned to that specific group. Since most of the patients received both a face-to-face and an e-consultation in 2020, for these patients a computerized random sample was derived from the study population by using Stata SE (version 15.0). According to Dutch guidelines, patients visit the rheumatologist every 3 to 4 months.

Data collection

Data were extracted from the Business Intelligence data warehouse of Maasstad Hospital, that holds financial data concerning all healthcare activities and procedures performed at Maasstad Hospital (i.e. in the rheumatology department and all other departments). Electronic health records were consulted to examine the number of patients receiving e-consultations and/or face-to-face consultations. Health costs were retrieved from the electronic medical dossiers of the patients, that provide information on the healthcare procedures performed and the related cost prices.

(Clinical) assessments

The patients' age, gender, disease duration, disease activity scores (DAS28CRP) and number of (multi-)morbidities were used to assess whether there was potential bias between the intervention and control group in terms of baseline characteristics. Disease duration is defined as the time (in years) between the diagnosis date and January 1st 2020. The number of multi-morbidities is stated as the occurrence of at least one other diagnosis besides RA. The DAS28CRP is a measure for the disease activity in RA patients by assessing the 28-joint count and the C-reactive protein (CRP) levels in the blood, the scores ranging from 0 to 9.4. A DAS28CRP-score below 2.6 indicates remission of RA [16].

Patient-reported outcomes

Outcomes, i.e. utilities, are obtained from PROM questionnaires which are filled out at home approximately two weeks before a consultation. At the rheumatology department of the Maasstad Hospital, the ICHOM standard set for Inflammatory Arthritis is implemented to assess the PROMs every 6 to 12 months as advised by ICHOM [14]. This standard set comprises five questionnaires of which, for the purpose of this study, the Work Productivity and Activity Impairment (WPAI), the Health Assessment Questionnaire - Disability Index (HAQ-DI), Rheumatoid Arthritis Impact of Disease (RAID) and the generic EQ-5D were assessed. The HAQ-DI is a score ranging from 0 to 3 and the scale of the RAID questionnaire runs from 0 to 10, with higher scores indicating worse health outcomes [17] [18]. These specific questionnaires were selected since they are most commonly applied in research and in clinical practice concerning RA [19] [20]. Moreover, the HAQ-DI and RAID are included as these questionnaires cover a broader and more RA specific range of patient-relevant domains, frequently used in healthcare cost studies [17] [18]. The difference between the RAID and HAQ-DI is that the HAQ-DI is specifically geared toward the physical functioning of patients, whereas the RAID comprises RA specific domains that are considered relevant to be patient. In comparison with the EQ-5D questionnaire, the RAID questionnaire encompasses the additional RA relevant patient domains of fatigue, sleeping problems and generally dealing with RA.

Costs

Healthcare costs as well as non-medical expenses were incorporated in the study. Healthcare costs include all expenditures incurred by the hospital, such as consultations, laboratory, inpatient, emergency and medication costs (see appendix 3) within one year from the baseline measure (first quarter of 2020). Regarding the non-medical costs, travel costs (direct) and productivity losses (indirect) were considered. The travel costs were only calculated for the conventional outpatient visit group, assuming that the intervention group (e-consultations) had no travel costs. Zip codes of participants were used to determine the distance from the patients' home addresses to the outpatient clinic at Maasstad Hospital in Rotterdam. The distance in kilometers for the required return trip was calculated per patient. Subsequently, the kilometers were multiplied by the assumed cost of €0.19 per kilometer and €3.00 per hospital visit was added with respect to the parking costs [21]. The productivity loss or lost wages applies to the study population under 67 years of age, as patients above this threshold are formally retired citizens who should not experience productivity losses. The loss of productivity regarding the visit to the hospital was estimated using the Human Capital method. The productivity losses were examined through the earlier mentioned WPAI questionnaire. Since the recall period concerning the WPAI questionnaire is one week, the potential productivity costs were only calculated for the week the consultation took place. To value the total productivity loss as a result of RA, the working hours lost were multiplied by the mean average wage per hour. The Dutch mean hourly wage is €36.40 according to Statistics Netherlands [22]. Costs were not discounted since the time horizon of the study was limited to one year and all costs calculations were based on 2019 prices.

Cost-effectiveness analysis

Decision tree

To examine the cost-effectiveness (utility) of telemonitoring concerning a societal perspective, a decision tree was constructed that consisted of two primary pathways.

A decision analytical tree method was selected given that two alternative forms of consultation, i.e. face-to-face and e-consultations, were evaluated. The results of the decision tree were used to evaluate the quality of life by assessing the HAQ-DI, RAID and EQ-5D questionnaires. The results of the health effects from the patient perspective were compared with the quality of life as expressed in QALYs. Depending on the random assignment, patients with both face-to-face and e-consultations in 2020 followed either the e-consultation branch or the face-to-face consultation branch. Patients can move to different health states depending on their DAS28CRP scores. The DAS28CRP scores were applied as cut-off values regarding the health states in the decision tree. A score of 2.6 or lower indicates RA remission and higher values correspond with an inflammatory (active) state of disease [23].

Different input parameters are required for the decision tree analysis. The utilities in the different health states are obtained from the RAID, HAQ-DI and EQ-5D questionnaires. The transition probabilities were calculated based on the real-world cohort, i.e., the number of patients moving to the specific health states. To estimate the cost-effectiveness of econsultations, an Incremental Cost Effectiveness Ratio (ICER) was calculated by subtracting the expenditures in the face-to-face consultation group from the expenditures in the e-consultation group and dividing the costs by the score of the EQ-5D, HAQ-DI and RAID respectively, for the intervention group versus the control group. A cost-effectiveness threshold of ϵ 50k per QALY was considered, which is in line with the relevant Dutch guidelines [21].

Statistical analysis

Descriptive statistics were used to summarize the baseline characteristics of the two patient groups. Depending on the data distribution, continuous variables are presented by their mean and standard deviation (unless stated otherwise) and categorical variables as percentages. Although most patients were randomly allocated between the two groups, the characteristics of the intervention group and control group were tested for potential bias since the consultation type was assessed retrospectively. The unpaired sample t-test and the Mann Whitney U test were applied to examine the statistical difference between the mean costs and health outcomes of both groups. Additionally, a sensitivity analysis with t-tests and Mann Whitney U tests were performed to assess the difference between patients who solely had e-consultations and face-to-face consultations. With respect to the HAQ-DI and RAID, the scores were converted into an EQ-5D equivalent scale to increase the comparability of the disease specific (RA) versus the generic health perspective. Following the approach applied in a previous study with respect to the HAQ-DI and RAID

scores were converted to a comparable EQ-5D score through linear regressions with EQ-5D scores as explanatory variable and the HAQ-DI or RAID as outcome variable. To test the generalizability of the outcomes for a RA population, the entire Maasstad Hospital patient population filling out PROMs in 2020 (N=890), instead of the applied study sample, was used as validation cohort.

Sensitivity analysis

To examine the robustness of the model results and to identify the key cost drivers, univariate and probabilistic sensitivity analyses were conducted. As part of the univariate sensitivity analysis, the healthcare perspective, i.e., excluding productivity losses, was selected to assess the switch of perspective on the ICER. A Monte Carlo simulation was conducted to perform the probabilistic sensitivity analysis, resulting in a cost-effectiveness plane. A gamma distribution was applied regarding the costs, as costs are generally skewed to the right, i.e., outliers with high expenditures, and a beta distribution was considered regarding the outcomes [25]. All data analyses were conducted using StateSE 15 for Windows, whereby a p-value of 5% was considered to be statistically significant.

Results

Patient population

A total of 772 patients were included from the 1231 patients who participated in the PROM inquiry, of which 148 patients solely had e-consultations and 41 patients only face-to-face consultations (table 1, appendix file 1 and 2). Approximately 60% of the included patients had available PROMs within the time frame of six months after the consultation date. The patients in the econsultation group were on average slightly younger (59.5; SD = 13.0) in comparison with the face-to-face consultation group (60.5; SD = 11.9). Females were overrepresented in the face-to-face consultation group (71.8%). Multimorbidities were slightly more prevalent in patients of the face-to-face consultation group and the mean disease activity (i.e. DAS28CRP) was lower in the e-consultation group were not statistically significant on a 5% alpha level. The sensitivity analysis concerning the patients with solely e-consultations and face-to-face consultations also showed no significant differences.

Table 1. Patient characteristics

| | | Face-to-face consultation (N=386) | E-consultation |
|--------------------------------------|--------|-----------------------------------|----------------|
| | | | (N=386) |
| Female (N, %) | | 277 (71.8) | 262 (67.9) |
| Age (years) | | 60.5 (11.9) | 59.5 (13.0) |
| Disease duration (years) | | 9.2 (4.6) | 8.8 (4.6) |
| Disease activity score (range 0-9.4) | | 2.6 (1.0) | 2.4 (1.0) |
| % Inflammation | | 41 | 45 |
| Presence of multi-morbidities | (N, %) | | |
| | 0 | 92 (23.8) | 105 (27.2) |
| | 1-5 | 219 (56.8) | 219 (56.7) |
| | 6-10 | 61 (15.8) | 52 (13.7) |
| | >10 | 14 (3.6) | 10 (2.6) |

Note. Data presented as mean and standard deviation unless stated otherwise

Decision tree analysis (outcomes and costs)

The probabilities of patients moving from one health state to the other, are depicted in table 2. For the patients in the face-to-face consultation group, the probability of inflammation is 41%. Hence, the probability of being in a state of remission is 59%. Regarding the e-consultation group, patients' chance of being in a state of remission is slightly lower than in the control group (55% versus 59%).

Table 2. Model outputs: health effects (patient-reported outcomes)

| Consultation | Disease activity status | HAQ-DI* | RAID* | EQ-5D¶ |
|--------------|-------------------------|------------|------------|-------------|
| type | | (mean, SD) | (mean, SD) | (mean, SD) |
| Face-to-face | Remission | 0.60 (0.6) | 3.13 (2.3) | 0.77 (0.19) |
| | Inflammation | 1.10 (0.7) | 4.32 (2.2) | 0.66 (0.26) |
| Electronic | Remission | 0.56 (0.6) | 2.64 (2.2) | 0.79 (0.18) |
| | Inflammation | 1.03 (0.7) | 4.62 (2.1) | 0.66 (0.24) |

Note. HAQ-DI: Health Assessment Questionnaire - Disability Index, RAID: Rheumatoid Arthritis Impact of Disease. Disease activity status (DAS28CRP): remission < 2.6, inflammation \ge 2.6; higher scores on RAID and HAQ-DI indicate worse outcomes. *p<0.05 in face-to-face group. \$p<0.05 in electronic group

Outcomes

The average HAQ-DI, RAID and EQ-5D scores are displayed in table 2. A lower score on the HAQ-DI and RAID scale indicates a better patient-reported health outcome, whereas for the EQ-5D the contrary applies. In the e-consultation group, patients in remission scored 0.56, 2.64 and 0.79 (mean, SD) on the HAQ-DI, RAID and EQ-5D respectively and for inflammatory patients this was 1.03, 4.62 and 0.66. Patients in remission in the control group had an average score of 0.60 (HAQ-DI), 3.13 (RAID) and 0.77 (EQ-5D), compared with inflammatory patients mean scores of 1.10, 4.32 and 0.66. Comparing the scores between the remission and inflammation patients by means of the Mann-Whitney test, showed that the inflammatory patients in the face-to-face consultation group had significant better outcomes on the RAID and EQ-5D scores (p<0.01), however patients scored worse on the HAQ-DI scores (p<0.01). The patients in remission within the e-consultation group (p=0.03), but not on the HAQ-DI (p=0.870) and EQ-5D scores (p=0.229).

Costs

Figure 1 illustrates the total costs incurred for RA patients, both by consultation type and health state. Overall, patients in the face-to-face consultation group induced higher costs, with cost savings for the e-consultation group of $\epsilon_{1,066}$ per patient. Differences in costs were predominantly because of direct healthcare expenses, consultations and medication costs and as a result of travel costs. However, the unpaired t-tests shows no significant difference in costs between the face-to-face and electronic group (p=0.225). The same was found for the patients in remission in the two groups (p=0.592). Inflammatory patients in the face-to-face group caused significantly higher expenditures than in the e-consultation group (p=0.041). In the latter group, 60% of patients had at least one face-to-face group that percentage was 96%.

Figure 1. Model outputs: costs (per patient)



Cost-effectiveness analysis

To compare the cost effectiveness of e-consultations versus face-to-face consultations, the patient-reported outcomes based on the RA specific measures HAQ-DI and RAID were converted into comparable generic measure EQ-5D scores. The equation 1 and 2 convert the HAQ-DI and RAID scores in EQ-5D values are displayed in equation 1 and 2 below. The transformed HAQ-DI values range from 0.889 in an optimal health state to 0.304 in the worst health state. Regarding the RAID score, 0.933 is the maximum EQ-5D equivalent value (perfect health) and the minimum value 0.383. The R² value of both equation estimates was 0.42, indicating that the RAID and HAQ-DI score explain 42% of the variability in EQ-5D measure (p<0.001). Regarding the robustness of the estimates, the complete PROM sample (N=890) generated similar estimates for both the HAQ-DI and RAID: 0.891 – (0.213 * HAQ-DI) with a R² value of 42% and 0.955 – (0.062 * RAID) also with a R² of 0.42.

Equation 1. Equation to convert HAQ-DI scores into EQ-5D scores EQ-5D=0.889-(0.195*HAQ-DI)

Equation 2. Equation to convert RAID scores into EQ-5D scores EQ-5D=0.933-(0.055*RAID)

The results of the cost-effectiveness analysis (CEA) are depicted in table 3. The e-consultation group scored 13 points more favorable on the conventional, i.e., unconverted, HAQ-DI questionnaire and 34 points better on the unconverted RAID questionnaire, the difference was 2.5 and 1.8 in terms of transformed QALYs respectively. Although the favorable difference per patient in the e-consultation group is small in terms of HAQ-DI and RAID scores (i.e. health outcomes), implementation of e-consultations is a cost-effective strategy; - ϵ 163,159 based on the HAQ-DI(QALY) and ϵ 223,002 for RAID(QALY). Regarding HAQ-DI and RAID scores, e-consultations are cost-effective given the Dutch QALY threshold of ϵ 50K. The same conclusion can be drawn when considering the conventional cost-utility analysis based on EQ-5D scores, as the implementation generates a cost saving of - ϵ 161,491 per QALY gained.

| | Costs | HAQ-DI | RAID | QALYs* |
|----------------------------|------------|--------------|--------------|------------|
| | | (QALYs) | (QALYs) | |
| E-consultations | €2,874,021 | 298 (285) | 1363 (285) | 285 |
| Face-to-face consultations | €3,285,435 | 311 (282) | 1397 (272) | 283 |
| Increment | - €411,414 | - 13 (2.5) | - 34 (1.8) | 2.6 |
| ICER | | - €31,816 | - €12,265 | - €161,491 |
| | | (- €163,159) | (- €223,002) | |

Table 3. Cost-utility analysis

Note. A higher score on the questionnaires is worse; therefore the effect is reversed; *obtained from the EQ-5D

Sensitivity analysis

A change from a societal to a healthcare perspective, affected the expenditures. The switch of perspective unaltered the health outcomes, however the difference in expenditures resulted in slightly lower cost savings, €949 instead of €1,066 from the societal perspective. This resulted in less negatives ICERs compared to the societal perspective. ICERs decreased from - €161,491 to - €143,750 for the EQ-5D, - €223,002 to - €198,504 for the RAID(QALY) and - €163,159 to -€145,235 with respect to the HAQ-DI(QALY) scores.

Results from the probabilistic sensitivity analysis indicate that in 37.8% (HAQ-DI), 27.3% (RAID) and 21.7% (EQ-5D) of the 1000 Monte Carlo iterations, e-consultations were less effective from a societal perspective, although they were cost saving (figure 2). In the majority of the probabilistic sensitivity analysis cases, the expenses of the e-consultations were lower and resulted in better health outcomes than face-to-face consultations.









Discussion

Integrating the disease specific RA patient perspective in an economic evaluation by means of the HAQ-DI and RAID is a promising alternative for the traditional generic measure EQ-5D. The implementation of e-consultations for RA patients is a dominant strategy according to economic evaluation considering both the EQ-5D and RA specific outcome measures. The ICER of the generic outcome measure, i.e. – \in 161,491 per QALY(EQ-5D) gained, corresponds to the ICERs encompassing the RA specific perspective, namely – \in 163,159 per HAQ-DI(QALY) gained and – \in 223,002 per RAID(QALY) gained in a RA cohort, treated in a secondary trainee center. Although the ICERs of the generic and RA specific measures are comparable, incorporation of disease specific patient-reported outcomes in economic evaluations have been proposed as method to better capture the patient-relevant domains of the disease impact than the generic measure EQ-5D. Hence, the use RA specific measures in cost-effectiveness analysis of healthcare technologies offers the opportunity to improve the assessment and treatment RA patients.

Although ICERs of both the RA specific reported outcome and generic outcome measures indicate that e-consultations are cost-effective, that effect is predominantly achieved through cost savings. A likely explanation for the cost savings is the lower probability of at least one (face-to-face) follow-up consultation for patients within the e-consultation group. Probably patients that are in remission and have a stable disease process and/or milder symptoms are less likely to opt for a face-to-face consultation. For those patients an e-consultation suffices.

The differences in outcomes of the cost-effectives analysis, based on (transformed) QALYs, are minor between the e-consultation and face-to-face group. These results are in line with other articles investigating the impact of telemonitoring on the cost-effectiveness [3] [26]. Because the HAQ-DI and RAID scores are converted to EQ-5D scores by performing linear regressions on a relatively small study population, the robustness of the results is potentially affected. However, similar approaches are conducted in the literature [24] [27]. A possible explanation for the difference in the ICERs of the HAQ-DI and EQ-5D versus the RAID, is the broader range of the RAID, capturing more RA patient-relevant domains. Furthermore, the EQ-5D and HAQ-DI questionnaires are both prone to ceiling effects, negatively affecting the sensitivity to changes in scores caused by increasing age and disease duration [29]. Hence, the RAID can be exploited as key PROM for RA patients with respect to the assessment of the disease specific relevant outcomes [30].

As healthcare systems are under increasing pressure regarding budgets, the evaluation of new technologies but also of the current healthcare delivery is of importance [31]. However, although the current Dutch guideline advises to apply EQ-5D and alternative questionnaires may only be used in addition to the reference case (i.e. EQ-5D) [21], the need to move beyond the classical viewpoint concerning the evaluation of healthcare interventions is increasing. As a result, several initiatives have been introduced, such as the ISPOR value flower [13]. The goal of the ISPOR value flower is to enhance a cost-effectiveness analysis in terms of the relevant aspects considered [13]. An example is the incorporation of the value of hope, referring to patients' risk tolerance, i.e. the value patients place on the variability in outcomes [13]. The concept of the ISPOR value flower is however geared toward evaluations from a more process and societal viewpoint instead of a patient centered approach. Therefore, examining PROMs can complement the ISPOR value flower, as data are assessed at patient level (i.e. micro level) in the shift toward more personalized care delivery.

The findings of this study should be interpreted considering certain potential limitations. First, the cohort of RA patients was followed during the COVID-19 pandemic, since in the preceding years, the utilization of e-consultations was too low to allow for a reliable cost-effectiveness analysis. However, the exceptional circumstances of the pandemic could bias the results, as for example the more severe or complex RA patients opted for face-to-face consultations. To address that bias, a random sample was created by placing patients in either the e-consultation or face-to-face consultation group. Based on the performed statistical tests regarding the baseline characteristics

of the two groups, a (weak) statistically insignificant difference was determined in terms of disease activity. The DAS28CRP scores were on average 0.2 points higher in the face-to-face consultation group compared with the e-consultation group, which can explain the higher expenditures and lower health outcomes in that group. A second limitation is related to the questionnaires, as nearly 40% of the patients filled out the questionnaires outside the recommended timeframe of 6 months. Therefore, the patient reported as well as generic outcomes will be less representative concerning the actual health outcomes at the time of the consultation. A third limitation arises in terms of the productivity costs, because the WPAI questionnaire is limited to the productivity losses in one week, a comprehensive view on the productivity losses over a longer time period is lacking. To minimize the impact on the research findings, productivity costs were exclusively calculated for the week in which the consultation took place. Due to the limited time frame concerning the WPAI questionnaire, the productivity losses are likely to be underestimated. Lastly, primary care costs were not considered in the ICER due to privacy laws impeding the data exchange of patients in the Netherlands. However as diagnosing, treatment and monitoring of RA takes place in the hospital, primary care costs are expected to be low.

Strengths of the study comprise the use of real-world data to examine the impact of econsultations on both costs and health outcomes, instead of obtaining utilities and transition probabilities from the literature. To our knowledge, this is the first time a comparison of the disease specific perspective with the generic perspective in a cost-effectiveness analysis for e-consultations by RA patients is made. The fact that a societal perspective is considered, is important for patient empowerment. A third strength concerns the robustness of the outcomes: the univariate and multivariate probabilistic sensitivity analysis of the results of the performed ICERs yielded comparable results. Furthermore, an internal validation was conducted to ensure that the obtained values from the conversion equations of the PROMs into the patient valued QALYs, were robust in a larger study sample.

To further enhance the inclusion of patient reported outcomes in economic evaluations, future research should focus on the creation of general conversion equations for disease specific outcomes by conducting the research on a broader patient population. In addition, the follow-up should be extended to examine the effects of electronic consultations in the long-term and inclusion of patient experience indicators as an outcome parameter could provide insights with respect to the delivered care.

7

Conclusions

In conclusion, to foster the incorporation of disease specific patient-reported outcomes in CEAs, this study provided evidence on the implementation of PROMs in an economic evaluation of RA consultations within a prevalent RA cohort, treated in a secondary single-hospital. Electronic consultations showed to be a cost-effective strategy in terms of scores on the RA specific measures RAID and HAQ-DI, comparable to the score based on the more generic EQ-5D. As PROMs are increasingly adapted in clinical practice, providing patient-relevant perspectives not reflected in generic measures, these tailormade PROMs will better reflect the patient domains in CEAs than generic outcome measures. Hence, as disease specific PROMs provide more information regarding the effects of treatment decisions, they are recommended for application in economic evaluations over generic measures.

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Appendix



Supplementary file 1. Study population selection

Supplementary table 1. Patient characteristics of the patients with one consultation type

| | Face-to-face consultation | E-consultation | |
|---|---------------------------|----------------|--|
| | (N=41) | (N=148) | |
| Female (N, %) | 28 (68) | 102 (69) | |
| Age (years) | 63 | 61 | |
| Disease activity score (range 0-9.4) | 2.75 | 2.69 | |
| Number of patients with multi-morbidities (N,%) | 21 (51) | 85 (57) | |

Note. Data presented as mean unless stated otherwise

| Costs categories | Examples |
|------------------|---|
| Diagnostics | X-rays and ultrasounds |
| Emergency | Ambulance, emergency hospitalization |
| Outpatient care | Consultations, procedure |
| Inpatient care | Operations, hospitalization |
| Medication | All medication provided by the hospital pharmacy e.g. |
| | (TS)DMARDS |
| Laboratory | All laboratory determinations e.g. CRP, ALAT, sodium |

Supplemental table 2. Healthcare costs included

7



Chapter 8

How Do Bundled Payment Initiatives Account for Differences in Patient Risk Profile? a Systematic Review

Hendriks CMR, Koster F, Cattel D, Kok MR, Weel-Koenders AEAM, Lopes Barreto D and Eijkenaar F

Submitted



Chapter 9

General Discussion

This thesis aims to contribute to the health economics literature by providing evidence for meaningful applications of value-based healthcare (VBHC) principles in a Dutch hospital setting. The underlying societal objective of this thesis is to establish and preserve a sustainable (i.e., affordable and equitable) healthcare system by aligning the viewpoint of patients with that of healthcare organizations and policymakers.

To address the issue of increasing healthcare costs, policymakers have attempted a myriad of policies over the last decades [1] [2]. However, within these policies, the patient perspective is often overlooked because policy emphasis is predominantly on cost containment. VBHC is frequently associated with quality, a concept that encompasses various dimensions. Within the context of healthcare, it places significant emphasis on inputs and adherence to processes. Should the patients' point of view already be considered, it is mostly limited to economic evaluations.

The basic principle behind an economic evaluation is to carefully analyze the costs and outcomes associated with each option under consideration [3]. This analysis helps to determine whether the investment in these options is valuable [3]. Economic evaluations suffice to reach health states for patients, such as progression free survival. However, progression free survival fails to adequately represent the quality of life as perceived by patients undergoing the treatment. Especially in the current era of rapidly evolving (technological) innovations, the patient experience and perception of care delivery is of increasing relevance to make more informed decisions regarding healthcare spending.

Over the past decades, a variety of VBHC initiatives have been initiated to maximize patient value per euro spent. However, due to the ambiguity in the definition of VBHC, the focus of the VBHC initiatives also deviates. Assuming that one of the key elements, such as leadership, integrated practice units and standardization of outcomes are present, VBHC can improve healthcare systems [4].

Although, the definition of VBHC varies widely in the literature, the core aspect, namely providing maximum patient value, is broadly agreed upon in the literature [4]. In that sense, VBHC offers a solution with respect to the incorporation of the patient perspective in terms of optimizing patient value in the decision making regarding clinical pathways. In this thesis, Porter and Teisberg's value agenda has been used as a guideline to research ways to implement VBHC in hospitals or other healthcare organizations and on a policy level [3]. The following paragraphs provide a discussion of the topics covered in the thesis chapters, along with the key findings.

1. Organize into integrated practice units

Organizing care delivery into Integrated Practice Units (IPUs) is the first step of the value agenda [5]. To facilitate the restructuring of healthcare services within the specialism of rheumatology based on the patient's medical condition, the care delivery value chain (CDVC) was used (chapter 2). The CDVC has been identified as the foundation for integrating VBHC into healthcare delivery, complementing evidence-based medicine and improving value by reorganizing care delivery on the meso and macro level. The development of the process map for Rheumatoid Arthritis (RA) patients by following a mixed method approach (i.e. a combination of quantitative and qualitative research) enables consistent analysis that can also be used as a template for other chronic illnesses. Notably in the transition from traditional care to care delivery with an IPU orientation, the CDVC will provide valuable insights to rearrange processes.

Implementing the CDVC in a real-world setting allowed to analyze the delivery of healthcare services for patients with RA in detail. As a result, inefficiencies were detected and processes aligned with the patients' needs. For instance, a telemonitoring track was suggested as part of the study. A patient panel was established to offer the possibility for rheumatology patients to co-decide or give advice on subjects relevant for these patients within the hospital. The patient panel gave advice regarding the design of the telemonitoring track.

2.1. Measure outcomes for every patient

Measuring outcomes is a key element of VBHC. However, integrating patient-reported outcomes into clinical care can be challenging [6]. Particularly when the tools required to measure and present these outcomes have to be incorporated into existing workflows without additional time constraints for healthcare professionals. Therefore, a shareddecision PRO-based dashboard was implemented at the rheumatology department of Maasstad Hospital and economically evaluated. Patients that used the dashboard in the consultation room with a rheumatologist had a higher quality of life enhancement and their healthcare costs were lower. Although this new way of working required some adjustment time, the initial study results were favorable.

By allowing for the evaluation of PROs in the consultation room, care provision is more personalized as in addition to clinical parameters, other relevant outcomes such as fatigue and mental wellbeing are topics of conversation. A positive association was found with respect to patients receiving assistance from the Joint Decision Support (JDS) dashboard and their scores on the HAQ-DI and the EQ-5D scores, as part of the PRO RA set. Patients utilizing the JDS dashboard experienced an average reduction of 14% in in-hospital healthcare costs compared to those who did not use the JDS dashboard in the consultation room.

There is a limited amount of research focused on the (quantitative) assessment of dashboard use in consultation rooms up to the present date. In other non-rheumatology disease areas, it was found that only a limited number of studies reported quantitative results on PROMs [7]. Out of these studies, four showed no significant effects on outcome improvements in terms of PROMs, while one study showed a minimal effect [7].

However, the use of dashboards enables the benchmarking of patients with specific medical conditions [8]. These so-called patients-like-me dashboards are implemented to enhance the quality of care from both a patient and healthcare organization perspective. Despite the recent developments in terms of PRO implementations, there are still several barriers to overcome before PROs can effectively be used on a health policy level [9]. Barriers involve the establishment of a standardized set of PROMs to be used as quality indicator and the ability to effectively translate these quality indicators into improvements in healthcare quality.

2.2 Measure costs for every patient

Time-driven activity-based costing (TDABC) has been proposed as the method to address the cost of creating and maintaining a model for cost allocation within VBHC [5]. TDABC offers a comprehensive and straight-forward method to model the cost framework of care delivery, enabling the identification of inefficiencies and cost-saving opportunities [10]. Cost-saving opportunities involve the treatment of the most complex (multimorbid) RA patients by a rheumatologist and other patients by less expensive medical staff members, such as physician assistants and nurse practitioners, as also concluded by other studies [11][12].

TDABC is however an advanced approach that requires substantial effort by researchers. TDABC has been suggested as a practical VBHC tool that extends beyond its role as a research method. It is important to note that implementing TDABC requires a considerable amount of effort from healthcare institutions to adopt and maintain the costing methodology. Consequently, TDABC is frequently limited to being conducted by researchers and not implemented in daily practice. However, to offer a less time-consuming method, Fuzzy-Logic (FL)-TDABC was applied in this thesis as a modification to further enhance the efficiency of VBHC-implementation. FL-TDABC empowers managers in healthcare to obtain more convenient access to the cost data. The data allows healthcare providers to be cognizant of the relationship between the costs and outcomes of medical treatments, a crucial aspect of VBHC [10]. Hence, it can guide providers in making well-informed decisions about investing in resources to enhance care quality and outcomes, while simultaneously managing costs [10].

By applying FL-TDABC in combination with a more efficient process of analyzing time estimates, cost accounting becomes accessible to a greater number of healthcare organizations. Hence, the actual costs of the care delivered to patients can be calculated relatively promptly as described in this thesis. The insights allow policymakers to evaluate the care delivery in a transparent and accurate manner as all the relevant costs are taken into account. In the present circumstances, cost data are not that patient centered, causing a lack of information concerning the time estimates of the trajectory.

3. Move to bundled payments

The movement towards bundled payments (BPs) as an alternative reimbursement model within VBHC involves challenging issues with respect to the design of BPs. As patients within bundles (care cycles) have different risk profiles, leading to variations in expected healthcare expenditures, accounting for these expected differences is of importance. To address the latter issue, a systematic review was conducted in the final chapter of this thesis. The most frequently used methods to address adverse incentives and minimize financial consequences of factors beyond providers' control are patient exclusions and risk adjustment of bundle prices. These methods primarily consider comorbidities, and then make adjustments for other factors such as sociodemographic, characteristics and condition-/procedure-specific factors.

According to Porter and Teisberg, BPs are the best suited payment model in accomplishing a VBHC system [5]. However, inadequate design of the BP in terms of the bundle definition and bundle price can lead to inequity, which conflicts with the principles of VBHC. This could result in care delivery being predominantly influenced by financial factors rather than patient needs and outcomes. Such a scenario may unwantedly restrict patients' access to essential care and potentially diminish the overall value provided by the healthcare system. Therefore, a sophisticated design of BPs is required to ensure access to healthcare now and in the future.

4. Integrate multi-site care delivery systems

To facilitate the integration of multi-site care delivery, the incorporation of the mesoand micro perspective in economic evaluations is needed. To augment from generic measures to assess healthcare services with elements that matter most to patients, PROMs provide a useful guidance. As PROMs represent the core of VBHC, they are particularly suitable to be incorporated in economic evaluations of for example treatments, services or vaccines. By applying PROMs in economic evaluations, the link between the patient perspective and the policy perspective is accomplished.

In this thesis, the generic questionnaire EQ-5D was replaced by the HAQ-DI and RAID, which are RA-specific PROs, with electronic consultations for RA patients as intervention. The outcomes showed that PROMs can be applied as an alternative outcome measure in economic evaluations, allowing for a more disease specific evaluation and monitoring of health outcomes. Due to the fact that such an assessment is possible, a broader perspective can be applied, incorporating not only the macro evaluation level, but also the micro and meso level. Furthermore, the integration of e-health through e-consultations improves communication across various care sites, providing patients with the opportunity to receive care from multiple locations rather than being limited to traditional healthcare settings.

5. Expand geographic reach

To expand the geographic reach and facilitate holistic care delivery, it is of utmost importance to understanding the differences in the health risk profiles of populations. Hence, a comparison was made between the expenditures within a single hospital and the Dutch (multimorbid) RA population to gain insight onto the degree and impact of these differences (chapter 4). It was found that in the context of RA, relying solely on the hospital perspective leads to an underestimation of the link between multimorbidity and healthcare costs, as 43% of healthcare utilization and expenditures remain unaccounted for. Another consequence for the outpatient clinic regarding multimorbid patients is the increased demand for mental health services, resulting in additional visits specifically as a result of mental complaints such as anxiety, depression and other psychiatric conditions.

To overcome this limitation and effectively treat multimorbid patients, healthcare professionals should provide holistic care and utilize data that offer a more comprehensive insight into patients' conditions. Enhancing the interoperability of data utilization among healthcare organizations will expand the geographic reach. Hence, expanding the geographic reach contributes to a sustainable value-based driven healthcare system.

6. Build an enabling information technology platform

An information technology platform facilitates the implementation of VBHC, where efficient IT tools support integrated and multidisciplinary care delivery [5]. Electronic

Health Records (EHRs) play a key role in providing information at the patient-level, which can be applied on a larger scale to assess costs and outcomes over a care cycle [13]. Nonetheless, misregistration occurs frequently. In chapter 5 the magnitude of the misregistration for RA was investigated including the financial consequences. At the rheumatology department of the Maasstad Hospital this occurred in around 19% of the RA diagnoses. Patients with misclassified diagnoses generate 10% higher yearly healthcare expenses, mostly driven by higher annual costs in diagnostics, outpatient, and inpatient care. Regardless of classification accuracy, patients reporting a better quality of life showed notably reduced healthcare costs. Hence, misclassification of diagnoses can negatively impact patient care and also influence the evaluation and comparison of health results and expenses.

Aligning IT tools with real-world data is described as the last fundamental component connecting all the above steps of the value agenda [5]. Lacking a supportive IT tools will potentially lead to inaccurate or incomplete information, having consequences on a micro, meso and macro level. On a micro level the patient will for example not receive the right treatment or care, on a meso level patients are compared with the wrong peers and on a macro level this could eventually lead to making inaccurate decisions with respect to the delivery of care. As a result, the quality-of-care decreases and the accessibility and affordability of care are eventually impacted as well.

Methodological constraints

The research conducted in this thesis is predominantly the result of analyses concerning real-world data. The data were retrieved from the RA population of Maasstad Hospital. Although this data comprehends a large set of RA patients, patients mostly originate from the metropolitan area of Rotterdam. This can influence the outcomes of the research, since the living conditions of patients in urbanized areas will differ from for example patients in more rural areas. Furthermore, Maasstad Hospital is located in the southern part of Rotterdam, in a neighborhood with a large population with a low socio-economic status. Hence, the results may not be generalizable to other populations. However, the data retrieved from Statistics Netherlands that were applied in chapter 5 revealed that the patients shared similar background characteristics.

An additional drawback of utilizing real-world data is selection bias. Selection bias indicates that the population incorporated in the study sample is not a true representation of the overall RA population [14]. For instance, patients that are included in the PROMs questionnaires query, might be the patients who for example experience higher or lower than average quality of life. In order to mitigate this drawback, patients are selected based on their diagnosis rather than relying solely on clinical and/or patientreported outcomes. Confounding is also a limitation of employing real-world data [15]. Confounding is the distortive relationship between an independent variable (exposure) and a dependent variable by the presence of a third variable, known as a confounding variable [15]. The confounder is linked to both the independent and dependent variable, which can result in an inaccurate association or a misinterpretation of the relationship between the two variables of interest [15]. The optimal research design regarding the data analyses incorporated is the randomized controlled trial (RCT) [16]. However, in case of an (economic) evaluation of two strategies such as in chapter 6 and 7, a RCT design was impracticable. Because of overlap, patients could not be blinded in groups engaging in face-to-face or electronic consultations, or that did or did not use a dashboard during the consultation. Moreover, real-world data serves as a representation of everyday practice. And as such, it does not require a sophisticated analysis such as a RCT, to be deemed trustworthy [17]. The primary emphasis of real-world data is on measuring effectiveness, while RCTs focus on assessing cost-effectiveness.

Another constraint of this thesis is related to the interoperability of the data. The data used were mostly limited to the RA patient of Maasstad Hospital, as data exchange on a patient level with other providers such as the GP or paramedical care providers is challenging due to privacy laws and the proliferation of IT platforms used in healthcare. Hence, an overall evaluation including primary, secondary, tertiary and other types of care was not conducted in this thesis.

Implications for future research

In the context of the Netherlands, healthcare challenges are often addressed by proposing reforms to the financing and reimbursement system. Currently, the Dutch healthcare system operates based on market actions, competition, and performance contracts [17]. However, completely overhauling the system through reform is a time-consuming and expensive process. Alternative systems, such as national health services or private healthcare systems, also have their drawbacks, including waiting lists and inequitable access to care [18][19]. In this regard, implementing VBHC offers a practical and efficient alternative.

To improve the sustainability of the Dutch healthcare system, it is crucial for the various subsystems to collaborate more intensively. By fostering cooperation and coordination among these subsystems, the overall effectiveness and efficiency of the healthcare system can be enhanced. This collaborative effort will contribute to the long-term viability of the Dutch healthcare system.

Although the body of thought concerning VBHC originates from 2006, the implementation of VBHC practices was delayed and occurred in the recent years [5] [20]. One challenge in adopting VBHC is the variation in its definition across healthcare settings and organizations, making comparisons difficult. The same holds for the comparison of this thesis to the literature. The movement to integrated practice units, the uniformity of patient outcomes and the incorporation of the patient perspective are considered as key elements in the literature and correspond with the findings of this thesis [4].

However, to overcome the challenges and successfully implement VBHC in practice, there are practical steps that can be taken, such as:

- Standardize the definition of VBHC: establish a clear and consistent definition of VBHC across healthcare settings and organizations to facilitate meaningful comparisons and benchmarking.
- Implement Patient-Reported Outcomes (PROs): utilize PROMs in the consultation room to measure outcomes at the individual patient level. These data can also be used to compare outcomes within specific disease areas or healthcare organizations, enabling benchmarking and identification of best practices.
- Extend benchmarking to a policy level: apply PROMs to compare treatments and outcomes on a policy level. This will allow for evidence-based decision-making and the identification of effective interventions.
- Adopt a value agenda: embrace the value agenda, which focuses on delivering high-quality care at an optimal cost. This involves aligning incentives, promoting transparency, and continuously improving processes and outcomes.

By following these practical recommendations, healthcare organizations can successfully implement VBHC and drive positive change in the healthcare system. This will ultimately lead to improved patient outcomes, enhanced efficiency and the long-term sustainability of the (Dutch) healthcare system.

Concluding remarks: a directive for value-driven healthcare enhancement

This thesis offers practical tools and guidance for implementing VBHC in real-world settings. It promotes a shift in clinical practice and costing system towards prioritizing patient value, which includes the consideration of quality of life and patient satisfaction with the healthcare delivery. When clinical practices are oriented towards patient-reported outcomes, not only is patient care enhanced, but the overall costs may be reduced by eliminating ineffective practices.

Rewarding qualitative good care measured through PROs

Healthcare providers should also be compensated based on the qualitative impact of their care, as measured by patient-reported outcomes. Alternative reimbursements strategies should be considered for those demonstrating improved patient-reported outcomes, which would reflect not just the medical success of treatments but also patient relevant factors of the care received. A value-based contract that Maasstad Hospital has concluded with the relevant healthcare insurers is a positive development in this direction.

In order to facilitate the transformation regarding this type of reimbursement and these type of contracts, economic evaluations should also be adapted. In addition to the generic outcomes, PROMs need to be evaluated and assessed in cost-effectiveness-and cost-utility analyses.

Reform in reimbursement strategies

The alignment of reimbursement strategies with patient outcomes is a critical lever in the reallocation of healthcare resources. It ensures that investments are directed towards services that deliver genuine value, as perceived by patients, which stands as a deterrent against wasteful expenditure and as a catalyst for the delivery of costefficient healthcare. Hence, the transition to a value-based reimbursement system may be achieved by implementing bundled payments that take into account the patient risk profiles of the within such a bundle (chapter 8).

The imperative of transparency for VBHC

For VBHC to realize its full potential, a climate of transparency concerning cost and outcome data is vital. The open exchange of such data is required, not merely for accountability but also to facilitate a culture of shared learning and quality enhancement across healthcare entities. It is this transparency that can drive a value-oriented transformation, leading to an effective containment of healthcare spending.

Need for economic sustainability in healthcare

The urgency to transition towards a value-based healthcare model is underscored by the pressing need to mitigate the unsustainable growth in healthcare costs. By incentivizing value over volume, healthcare providers are motivated to prioritize improved outcomes along with cost awareness, thereby safeguarding the sustainability of healthcare systems.

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Summary

The thesis aims to contribute to the health economics literature by providing evidence for meaningful applications of value-based healthcare (VBHC) principles in a Dutch hospital setting. It aligns the viewpoint of patients with that of healthcare organizations and policymakers, emphasizing the incorporation of the patient perspective in decisionmaking regarding clinical pathways.

Patient level (micro)

On a micro level, a validated process map of the care cycle for Rheumatoid Arthritis (RA) patients is established as a first step aiming to increase transparency and optimize care delivery. It uses the Care Delivery Value Chain (CDVC) approach to document activities and resources systematically, resulting in improvement actions on outcome and process levels. To link costs to the care delivery pathway of RA patients, time-driven activity-based costing (TDABC) is applied. TDABC is supported by fuzzy logic (FL) to estimate the annual costs and cost drivers of the RA care cycle. The FL-TDABC methodology offers a more precise and efficient estimate of care cycle costs, allowing for the subjective nature of healthcare time estimates made by medical staff.

Healthcare organization level (meso)

From a healthcare organization level, this thesis analyzes the magnitude and implications of misclassification within RA care from a clinical and financial perspective. Misclassification of RA diagnoses affects healthcare expenditures and the quality of patients' lives negatively, highlighting the need for accurate registration of diagnoses.

In addition, the prevalence of multimorbidity among RA patients is assessed, showing that a single-hospital approach underestimates the association between multimorbidity and healthcare expenditures. The study suggests that professionals need to use data providing comprehensive pictures of patients to efficiently coordinate multimorbid patients.

To apply VBHC in practice, dashboards depicting patient-reported outcomes (PROs) used in the consultation room are expected to enhance patient-centered care delivery. This thesis demonstrated that a PRO-integrated dashboard results in beneficial outcomes on PROs and expenditures.

Health policy level (macro)

A macro level evaluation in this study assesses the possibility to use diseasespecific measures in a cost-effectiveness analysis (CEA) of electronic consultations (e-consultations) compared with face-to-face consultations. Disease-specific patientreported outcome measures (PROMs) offer a promising alternative for traditional measures in economic evaluations, capturing patient-relevant domains more comprehensively. Furthermore, a systematic review of literature on bundled payments (BPs) initiatives was conducted to understand how differences in patient risk profiles (PRP) are accounted for. The review finds that BPs use a variety of approaches to account for differences in PRP, with room for improvement in design choices.

Portfolio

Name: Fiona Koster

PhD period: April 2020 – December 2023

Department: Rheumatology and clinical immunology, Maasstad Hospital Rotterdam Erasmus School of Health Policy and Management, Erasmus University Rotterdam

| PhD training | Year | Workload |
|--|--------|----------|
| Courses | | (ECTS) |
| Conducting an Intervention Review | 2020 | 1.0 |
| How to finish your PhD in time | 2020 | 2.5 |
| Data analysis in R-practical hiostatistics | 2020 | 2.5 |
| ICH-GCP: good clinical practice | 2020 | 0.3 |
| Brush un your research design | 2.02.0 | 2.5 |
| English academic writing for PhD students | 2020 | 2.5 |
| Value-based healthcare from theory to implementation | 2021 | 0.7 |
| Professionalism and integrity in research | 2021 | 1.0 |
| Multi-level modelling, an introduction | 2021 | 2.5 |
| Salf presentation, focus, structure interaction and visualization | 2021 | 2.5 |
| Value hased healtheare valley, halt | 2022 | 2.5 |
| value-based nearthcare yellow belt | 2023 | 1.0 |
| <u></u> | | 18.5 |
| (Inter)national conferences | | |
| Dutch Rheumatology days | 2021 | 1.0 |
| ICHOM (virtual) | 2021 | 1.5 |
| American College of Rheumatology (virtual) | 2021 | 1.0 |
| ISPOR Europe (virtual) | 2021 | 1.0 |
| Dutch Rheumatology days | 2022 | 1.0 |
| ICHOM | 2022 | 2.0 |
| ISPOR Europe (virtual) | 2022 | 1.0 |
| | | 8.5 |
| Seminars, workshops and master classes (e.g. at department) | | |
| Symposium Different Perspectives on DMARD-treatment strategies in RA | 2021 | 0.1 |
| Symposium Shared Decision Making | 2021 | 0.2 |
| Symposium Shared Decision Making | 2022 | 0.2 |
| | | 0.5 |

International peer-reviewed publications (published or preparing for submission)

| Subility | | |
|---|------|-----|
| Koster, F., Lopes Barreto,D., Nair, S.C. et al. Defining the care delivery | 2023 | |
| value chain and mapping the patient journey in rheumatoid arthritis. | | |
| Rheumatol Int 43, 743–750 (2023) | | |
| Koster, F., Kok, M.R., van der Kooij, J. et al. Dealing with Time Estimates in | 2023 | |
| Hospital Cost Accounting: Integrating Fuzzy Logic into Time-Driven Activity- | | |
| Based Costing. PharmacoEconomics Open (2023) | | |
| Koster F., Bakx P.L.H., Kok M.R., Barreto D.L., Weel-Koenders A.E.A.M. | 2023 | |
| Multimorbidity status and annual healthcare expenditures of rheumatoid | | |
| arthritis patients: a Dutch hospital-centered versus population-based | | |
| comparison. Rheumatol Int. 2023 Jun;43(6):1067-1076 | | |
| Koster, F., Kok, M.R., Lopes Barreto, D. and Weel-Koenders, A.E.A.M., | 2024 | |
| Capturing Patient Value in an Economic Evaluation. Arthritis Care Res. | | |
| Arthritis Care & Research, 76(2), 191-199. | | |
| Koster F, Dikkenberg van den M, Kok MR, Lopes Barreto D, Weel-Koenders, | 2024 | |
| Evaluating the implementation of a patient-reported outcomes dashboard: effects | | |
| on health expenditures and outcomes. Submitted | | |
| Koster F, Kok MR, Kok MR, Conijn C, Weel-Koenders AEAM, Lopes Barreto | 2024 | |
| D. Financial Impact of Clinical Misregistration in Rheumatoid Arthritis | | |
| Patients. Submitted | | |
| Hendriks CMR., Koster F, Cattel D, Kok MR, Weel-Koenders AEAM, Lopes | 2024 | |
| Barreto D, Eijkenaar F. How do bundled payment initiatives account for | | |
| differences in patient risk profile? a systematic review. Submitted | | |
| Dutch publications | | |
| 'Kosten zijn ondergeschoven kindje waardegedreven zorgonderzoek, maar | 2021 | |
| zó belangrijk." Publication at linnean.nl | | |
| 'Het belang van kosten in Value-Based Healthcare" Publication at | 2022 | |
| maasstadziekenhuis.nl | | |
| Waardegedreven zorg schuurt tegen het Nederlandse stelsel" Publication at | 2023 | |
| zorgvisie.nl/qruxx.nl | | |
| Presentations | | |
| Rheumatology research symposium | 2021 | 0.1 |
| Santeon VBHC Wetenschap Bijeenkomst | 2021 | 0.1 |
| Autumn Days of the Dutch Association for Rheumatology conference | 2021 | 0.5 |
| (podium presentation) | | |
| 'Science day" at Maasstad Hospital (poster presentation) | 2021 | 0.1 |
| Linnean Institute VBHC & costs conference | 2021 | 0.2 |

| ICHOM (podium presentation) | 2021 | 0.5 |
|---|-------|------|
| Autumn Days of the Dutch Association for Rheumatology conference | 2022 | 0.1 |
| (poster presentation) | | |
| "Science day" at Maasstad Hospital (podium presentation) | 2022 | 0.1 |
| ISPOR Europe (poster presentation) | 2022 | 0.2 |
| | Year | |
| | | 1.5 |
| Teaching | | |
| N/A | | |
| Lectures | | |
| Guest lecture VBHC course (BSc course) | 2022 | 0.2 |
| | | 0.2 |
| Tutoring, mentoring | | |
| Technology and innovation in healthcare (BSc course) | 2021 | 0.7 |
| Technology and innovation in healthcare (BSc course) | 2022 | 0.7 |
| | | 1.4 |
| Supervising | | |
| MSc thesis: "The effect of telemonitoring on time-driven activity-based | 2021 | 1.5 |
| costing in the management of inflammatory arthritis patients" | | |
| BSc thesis: "Supporting Shared Decision Making with a dashboard in | 2022 | 1.0 |
| Rheumatoid Arthritis Care" | | |
| MSc thesis: "Cost-effectiveness of electronic consultations versus face-to- | 2022 | 1.5 |
| face consultations in patients with rheumatoid arthritis | | |
| MSc thesis: "Economic evaluation of electronic consultations for Dutch | 2022 | 1.5 |
| rheumatoid arthritis patients by using value based healthcare quality | | |
| adjusted life years" | | |
| | | 5.5 |
| Other | | |
| Achievements | | |
| Research grant Maasstad hospital | 2021 | |
| Best new investigator poster research presentation award at ISPOR Europe | 2022 | |
| conference | | |
| Work experience | Start | End |
| Linnean Institute: member of workgroup costs | 2021 | 2022 |
| Santeon Research Network on creating value: organizing meetings | 2021 | 2022 |
| Early Career Network for Health Outcomes Research and Management: | 2022 | 2023 |
| treasurer | | |

Portfolio

About the author

Fiona Koster was born on March 1, 1993, in Den Helder. After living in the United States for a few years, she obtained her pre-university (Atheneum) diploma in 2012 at the Lyceum aan Zee. During her high school years, she spent 5 to 6 days a week in sports halls across the country, playing basketball.

After high school, she moved to Leiden to start a study in bio-pharmacy. It quickly became apparent that this was not the right choice for her, so she switched to health sciences at the Vrije Universiteit Amsterdam. In her third year of health sciences, Fiona was introduced to the field of health economics. After completing a minor in economics at the VU and participating in the Erasmus+ program at the Athens University of Economics and Business, she completed her master's in health economics at Erasmus University Rotterdam in 2018.

After a detour at an IT consultancy firm, she decided to return to the academic world and pursue a PhD at the Maasstad Hospital and as an external researcher at the Erasmus University Rotterdam. This was a practical-oriented research position with ample room for personal input. Currently, Fiona works as a health economic consultant at PwC.

Acknowledgements

Wie had ooit gedacht dat ik, na blijven zitten in 4 VWO en een officiële waarschuwing van de leerplichtambtenaar, uiteindelijk zou promoveren? Voor degenen die mij kennen, zal dit geen verrassing zijn. "Fiona doet alles op haar eigen manier" was de oneliner die ik te horen kreeg bij het behalen van mijn VWO-diploma.

Dit proefschrift zou echter niet tot stand zijn gekomen zonder de steun en bijdrage van alle mensen om mij heen. Daarom wil ik graag een aantal mensen, kort maar krachtig, bedanken voor hun onmisbare hulp en steun tijdens dit traject.

Als eerste wil ik mijn promotor Angelique bedanken. Jij hebt mij geleerd om sterk te staan in een academische omgeving en gaf mij veel vertrouwen als beginnend onderzoeker. Op de NVR, waar de reuma collega's mij op het feest en diner naar binnengesmokkeld hebben, hebben we de hele avond gedanst op ABBA. Maar het absolute hoogtepunt was toch wel het ICHOM congres, waar we, naast veel inspiratie, het nachtleven van Boston op z'n kop hebben gezet.

Deirisa, bedankt voor je steun de afgelopen jaren en je waardevolle input en constructieve feedback tijdens de beoordeling van mijn onderzoek. Jouw creativiteit op het gebied van onderzoek heeft mij vaak geënthousiasmeerd. Marc, dank je wel dat ik altijd jouw kantoor mocht binnenstormen om te klagen over de bureaucratie van het ziekenhuis of het feit dat ik beledigd was dat ik ergens weer geen toestemming voor had. Jij hebt mijn ongeduldige karakter altijd getolereerd en dit omgezet naar positieve energie.

Ik wil graag de commissie bedanken voor de beoordeling van mijn proefschrift: prof. Ahaus, prof. Boonen, dr. Van den Hout, dr. Koppert, prof. Stiggelbout, prof. Van der Nat en prof. Uyl-de Groot.

Graag wil ik alle coauteurs bedanken voor hun waardevolle feedback, zonder jullie was dit proefschrift er nooit gekomen. Ik ben ook dankbaar voor het Linnean Initiatief waar ik onderdeel mocht zijn van een werkgroep.

Daarnaast wil ik alle collega's en patiënten van de reumatologieafdeling van het Maasstad bedanken. En in het bijzonder Tessa, die mij in de lockdown alle 'ins and outs' van het Maasstad heeft geleerd. Maar ook Daisy, met wie we vaak met zijn drieën, waarbij ik op de prullenbak zat, geluncht hebben "achter de kast". Juul en Marijke, onze Whatsapp groep "Mentale Support Groep" zegt genoeg, net als de ruim 600 foto's die we naar elkaar gestuurd hebben. Martijn ook jij dank voor alle ondersteuning tijdens dit traject. Celine en Maud, vanuit ESHPM met wie de ESHPM wielrengroep is ontstaan. Het aantal keren dat we met z'n drieën gefietst hebben, is afgerond welgeteld nul.

Free Willy, ondanks dat biofarmacie niet helemaal mijn ding was, ben ik erg blij dat ik jullie ontmoet heb. De reis naar de VS het afgelopen jaar was wat mij betreft het absolute hoogtepunt waar we ontzettend veel hebben gelachen, karaoke gezongen hebben tot diep in de nacht, 4th of July hebben gevierd the American way en een wedstrijd van de Yankees hebben bezocht. Annelot, Julie en Meike kunnen ook niet ontbreken in dit dankwoord. Het was met jullie in de bunker van de VU dat ik interesse kreeg in gezondheidseconomie en dat Annelot en ik besloten om een master in Rotterdam te doen. Eenmaal op de EUR werden Annelot en ik vaak in een zin genoemd, bij hoge uitzondering mocht Thijs wel eens aanhaken tijdens de pauzes.

Daarnaast wil ik alle huisgenoten van de Korevaar bedanken. De Korevaar ofwel zwevende bank, het mooiste huis in Leiden, waar we helaas nooit een lustrum hebben mogen vieren. Toch waren jullie een belangrijk onderdeel van mijn studentenleven waar ik mij altijd thuis heb gevoeld. Op de huisdiners, de hifi- en tentavonden hebben wij oneindig veel avonturen beleefd. Van in je badjas naar de supermarkt, debatteren op huisavond, met waterpistolen van het dakterras schieten tot op vrijwillige basis thuisgebracht worden door de pendelbus of zelfs de politie.

Lieve beestjes, zonder jullie en met zo nu en dan "work hard, play hard", had ik het nooit gered. Hoewel het work hard gedeelte in het begin van onze studententijd niet echt hoog op prioriteitenlijst stond, wat ook wel duidelijk werd toen we officieus de vrouwen jaarclub waren die het meest aanwezig was op de tent. Ik ben ik heel erg dankbaar voor het feit dat wij nog steeds superhecht zijn. Ik hoop op nog heel veel mooie reizen (wellicht zonder een buddysysteem), borrels en feestjes met jullie!

Het sterrenstelsel, oftewel stars en mars, we kennen elkaar iets langer dan twee jaar. Dat zou je aan het aantal photobooth foto's die bij Sophie en mij op de Sloterkade hangen in ieder geval niet zeggen. Jullie zijn super meegaand en ik kan altijd op jullie rekenen, iets waar ik erg dankbaar voor ben! Dit geldt natuurlijk ook voor alle andere "glitter" meiden, op nog vele dansjes met jullie. En in het speciaal wil ik Sophie bedanken waarmee ik al anderhalf jaar in ons fijne appartement in Amsterdam woon, een fijner huis en huisgenoot kun je je niet wensen. Daarnaast wil ik ook Frank bedanken voor alle wijnavonden waarin we alle maatschappelijke problemen bespreken en ik altijd kan rekenen op een luisterend oor. Jan voor het zijn van mijn beste collega met wie ik vaker buiten kantoor dan op kantoor afspreek. En Youri voor het feit dat ik jou altijd kan bellen en je in mij gelooft.

Onze eerste ontmoeting was in de kleuterklas maar daar kunnen wij ons beiden vrij weinig van herinneren. Maar het moment dat ik terugkwam uit de VS werden wij meteen vriendinnen. We gingen naar dezelfde middelbare school en doopten onszelf om tot Fionica. Maar naast Monica ben ik ook dankbaar voor mijn andere middelbareschool vrienden, Hannelore, Ricardo en Robin.

Fay en Mies, onze vriendschap startte in de brugklas en bijna 20 jaar later zijn jullie nog steeds in mijn leven. Ik waardeer de oneindige woordgrapjes, tripjes naar het buitenland en wijnavonden. We hebben het nog vaak over de middelbare-schooltijd waarin wij de boel op stelten hebben gezet. Van containers voor de ingang van school plaatsen tijdens de examenstunt, elkaars naam doorgeven bij het te laat komen en via het bestuur van de school toch nog geld ontfutselen voor een evenement wat wij organiseerden.

Mijn paranimfen Mary en Louise wil ik in het bijzonder bedanken voor alle hulp. Als huisgenoten in Leiden hebben wij samen gelachen en gehuild en hebben wij ons ontwikkeld tot de personen die wij vandaag zijn. Lou ik waardeer het dat je altijd zo lekker eerlijk en oprecht bent en dat ik dankzij jou de uitspraak "het is een investering" heb geleerd, als we weer eens een paar schoenen of sieraden kochten. En Mar, geregeld elkaars plus een op feestjes, diegene met wie ik bijna dagelijks bel, die altijd een souvenir meeneemt, waarmee ik al meer dan 10 jaar naar Ajax ga en ik altijd om (ongezouten) advies kan vragen.

Jesse mijn "kleine" broertje of tweelingbroer zoals soms wel eens gedacht werd. Qua innerlijk zijn de gelijkenissen wat minder zichtbaar, wat onze gesprekken des te leuker maakt. Uiteraard geldt dit ook voor de rest van de familie Koster, veel dank voor jullie interesse!

En last but not least, mijn ouders. Het feit dat ik een combinatie van jullie ben, niet alleen qua genen maar ook qua studie en carrière, zegt al genoeg. Dankzij jullie heb ik een brede interesse, iets waar ik erg dankbaar voor ben. Door jou Helen, heb ik geleerd dat niet iedereen dezelfde kansen krijgt in de maatschappij en dat solidariteit belangrijk is. En Rob, wat hebben wij eindeloos gebeld over onze proefschriften. Onder het mom gedeelde smart, is halve smart. Zonder jouw input, had ik hier nooit gestaan. Je hebt mij echt in alle facetten van het onderzoek geholpen, dank je wel voor alle inspiratie, discussies en feedback.