Disinvestment Decisions in Healthcare

An exploration of mechanisms and considerations

Adriënne H. Rotteveel

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Disinvestment decisions in healthcare
An exploration of mechanisms and considerations

Beslissingen om de vergoeding van zorg stop te zetten
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(met een samenvatting in het Nederlands)

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Promotoren:

Prof. dr. K.G.M. Moons
Prof. dr. N.J.A. van Exel

Copromotoren:

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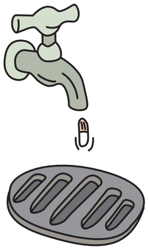
Outline

1

General Introduction



In many countries, healthcare expenditures have been increasing faster than the Gross Domestic Product (GDP) due to demographic developments, innovation and changes in the demand for healthcare [1, 2]. As a result, an increasing proportion of the GDP is spend on healthcare. This increases the pressure on the public financing of healthcare [1, 2]. To ensure that we can still afford our healthcare expenses in the future, the growth in healthcare expenditure needs to be contained.

In the past, policymakers have taken several policy measures to contain the growth in healthcare expenditures [3]. One of such policy measures is the strict assessment of new healthcare interventions that may qualify for reimbursement. With healthcare interventions we mean all types of curative care, such as medicines, therapies and medical devices. In the Netherlands, healthcare interventions are automatically reimbursed, except for outpatient pharmaceutical care, expensive inpatient pharmaceutical care and healthcare interventions for which there is no consensus on their effectiveness (e.g. that are not included in medical guidelines). The healthcare interventions that are not automatically reimbursed are, first, critically appraised by the National Healthcare Institute based on the formal assessment criteria effectiveness, cost-effectiveness, necessity and feasibility (see Table 1). Secondly, based on this assessment, the National Healthcare Institute advices the Dutch Ministry of Health, Welfare and Sports on the reimbursement of the healthcare interventions. Finally, the Minister decides whether the healthcare interventions are reimbursed [4-6].

Because of the focus of reimbursement decision-making on outpatient and expensive inpatient pharmaceutical care, most examples of the strict appraisal of healthcare interventions for reimbursement in the Netherlands are from the pharmaceutical field. For instance, there has been much debate on the reimbursement of expensive, innovative medicines for cancer and the reimbursement of expensive orphan drugs for rare diseases. Nevertheless, as healthcare interventions for which there is no consensus on their effectiveness are also assessed before reimbursement, there are also examples of other types of healthcare interventions which have been denied reimbursement. Such an example is knee-distraction for end-stage arthrosis of the knee, which was denied reimbursement because of insufficient evidence of effectiveness.

Previous studies have shown that effectiveness is an important criterion for the reimbursement of healthcare interventions in many countries [7, 8]. In the Netherlands, effectiveness even is a knock-out criterion, implying that healthcare interventions should not be reimbursed if they are not proven to be effective [5]. Despite of this, studies have estimated that, in OECD-countries, 20 to 30% of the healthcare budget is spend

Table 1: Formal criteria for reimbursement in the Netherlands [4-6]

Criterion	Description
Effectiveness (knock-out criterion)	Is there sufficient evidence of the effectiveness of the healthcare intervention?
Cost-effectiveness	Are the costs of the healthcare intervention acceptable relative to its effects?
Necessity	Consists of two criteria: 1) Is the disease at which the healthcare intervention is targeted severe enough? 2) Is the healthcare intervention expensive to such a degree that patients cannot pay for it themselves?
Feasibility	Is it feasible to reimburse the healthcare intervention? This, among others, concerns the organization of healthcare, administrative burden and ethical aspects. For all aspects of feasibility, see Couwenbergh et al. [4].

on ineffective healthcare interventions [9, 10]. For this reason, much can be gained, both in terms of improving health and containing healthcare expenditures, from the reassessment of healthcare interventions that are currently being used and being reimbursed in the healthcare system. The structured, evidence-based reassessment of healthcare interventions based on their clinical, economic, social and ethical aspects has been called Health Technology Reassessment (HTR) [11, 12]. In the Netherlands, it is assumed that when healthcare interventions do not meet the formal assessment criteria (anymore), they will automatically no longer be performed [4]. Thus, indicating that formal HTR is not needed. However, research has shown that ineffective healthcare interventions are not automatically no longer provided [13]. This indicates that formal HTR may be necessary after all.

One may argue that if HTR shows that a healthcare intervention does not meet the requirements for reimbursement, this healthcare intervention should no longer be reimbursed. The full or partial withdrawal of reimbursement of healthcare interventions by means of a policy decision has been called disinvestment [14, 15]. Despite of the potential gains from HTR and subsequent disinvestment, research has shown that policymakers generally consider stopping the reimbursement of healthcare interventions much more difficult than not reimbursing the same healthcare interventions in the first instance [16]. Even in cases where there was compelling evidence of ineffectiveness, proceeding to disinvestment was shown to be very difficult [12]. Unsurprisingly, a study reviewing empirical evaluations of disinvestment initiatives from many countries, concluded that only a limited number of these initiatives could be considered successful

[17]. In the Netherlands, the disinvestment of healthcare interventions is societally sensitive, with disinvestment often receiving limited societal support. For instance, the advices of the National Healthcare Institute for the disinvestment of the orphan drugs for the rare diseases of Fabry and Pompe in 2012, and a lung rehabilitation treatment for Asthma patients in Davos, Switzerland in 2019, received very limited societal support. In some cases, the lack of societal support for disinvestment decisions even resulted in the reversal of these decisions.

Summarizing, despite of the urgency and potential gains of disinvestment, disinvestment has been shown to be challenging. Specific mechanisms and considerations may be at play in disinvestment decisions that limit the feasibility of these decisions. Therefore, the objectives of this thesis are 1) to obtain insight in the mechanisms and considerations that are relevant in disinvestment decisions, and 2) to investigate aspects affecting the feasibility of these decisions. This information can be used to obtain more insight in the perceived difficulty of disinvestment decisions and provides guidance for a potentially more successful approach to disinvestment.

In **Chapter 2,** we study the disinvestment process in the Netherlands based on five recent cases of disinvestment. This chapter provides insight in the mechanisms underlying disinvestment processes and the aspects determining the outcome of these processes.

Chapter 3 describes a Q-methodology study that aimed to obtain insight in the societal viewpoints on disinvestment in the Netherlands. **Chapter 4** proceeds with describing the societal support for the viewpoints identified in Chapter 3. Moreover, it describes whether and how the support for the viewpoints is associated with background characteristics of citizens. The information obtained in these two chapters can be used to provide insight in why disinvestment decisions receive limited societal support and to assess whether there are approaches to disinvestment that may receive broader societal support.

Chapter 5 reviews the disparity between the willingness-to-accept (WTA) and the willingness-to-pay (WTP) for healthcare goods and services. WTA is the applicable criterion for disinvestment decisions as it weights losses in healthcare interventions against the savings resulting from these losses. WTP weights gains in healthcare interventions against their costs and is generally used in the context of new healthcare interventions that may qualify for reimbursement. Therefore, this systematic review and meta-analysis provides insight in whether losses in healthcare are valued differently from gains in healthcare. If losses in healthcare are indeed valued differently from gains, this may provide a possible explanation of the difficulty to disinvest healthcare interventions.

Chapter 6 describes a participatory value evaluation (PVE) study that aims to investigate the preferences of Dutch citizens towards the relative importance of attributes of healthcare interventions in the context of disinvestment. In this way, Chapter 6 provides further insight in the considerations the public find relevant in the context of disinvestment.

Finally, **Chapter 7** summarizes the findings of previous chapters, discusses the main conclusions of this thesis, lists a number of recommendations for future research, and discusses the potential implications of the Covid-19 pandemic on disinvestment decisions in the future.

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2

What Influences The Outcome Of Active Disinvestment Processes In Healthcare?

A qualitative interview study on five recent cases of active disinvestment

Rotteveel AH
Lambooij MS
van de Rijt JJA
Van Exel NJA
Moons KGM
De Wit GA

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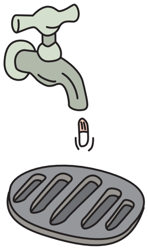

Abstract

Background: Recent attempts of active disinvestment (i.e. withdrawal of reimbursement by means of a policy decision) of reimbursed healthcare interventions in the Netherlands have differed in their outcome: some attempts were successful, with interventions actually being disinvested. Other attempts were terminated at some point, implying unsuccessful disinvestment. This study aimed to obtain insight into recent active disinvestment processes, and to explore what aspects affect their outcome.

Methods: Semi-structured interviews were conducted from January to December 2018 with stakeholders (e.g. patients, policymakers, physicians) who were involved in the policy process of five cases for which the full or partial withdrawal of reimbursement was considered in the Netherlands between 2007 and 2017: benzodiazepines, medication for Fabry disease, quit smoking programme, psychoanalytic therapy and maternity care assistance. These cases covered both interventions that were eventually disinvested and interventions for which reimbursement was maintained after consideration. Interviews were transcribed verbatim, double coded and analysed using thematic analysis.

Results: The 37 interviews showed that support for disinvestment from stakeholders, especially from healthcare providers and policymakers, strongly affected the outcome of the disinvestment process. Furthermore, the institutional role of stakeholders as legitimized by the Dutch health insurance system, their financial interests in maintaining or discontinuing reimbursement, and the possibility to relieve the consequences of disinvestment for current patients affected the outcome of the disinvestment process as well. A poor organization of patient groups may make it difficult for patients to exert pressure, which may contribute to successful disinvestment. No evidence was found of a consistent role of the formal Dutch package criteria (i.e. effectiveness, cost-effectiveness, necessity and feasibility) in active disinvestment processes.

Conclusions: Contextual factors as well as the possibility to relieve the consequences of disinvestment for current patients are important determinants of the outcome of active disinvestment processes. These results provide insight into active disinvestment processes and their determinants, and provide guidance to policymakers for a potentially more successful approach for future active disinvestment processes.

Background

Over the last decades, OECD countries have spent an increasing proportion of their gross domestic product (GDP) on healthcare [1-3]. It is expected that healthcare expenditures will keep growing faster than the GDP due to demographic changes and the introduction of technological innovations, increasing the pressure on public financing [3, 4]. To limit this increase in healthcare spending, governments worldwide have introduced cost-saving policy initiatives, such as lean thinking and retracting inefficient processes [5]. Furthermore, proposals for disinvestment (i.e. withdrawal, restriction, retraction or replacement [6, 7]) of healthcare interventions and services have been made [5, 8, 9]. Reasons for disinvestment may be, among others, ineffectiveness, unsafety or a lack of value for money [10, 11]. Furthermore, disinvestment can either be passive (i.e. not dependent upon direct policy intervention) or active (i.e. requiring direct policy intervention, such as a reimbursement decision) [12].

In the Netherlands, adults are obliged to buy individual private health insurance. Although this health insurance is provided on a competitive health insurance market, the coverage (i.e. the basic benefits package) is similar for everyone [13]. Most types of curative care are automatically included in the basic benefits package, except for outpatient pharmaceutical care and expensive inpatient pharmaceutical care. For these two types of care, assessment and appraisal by the National Healthcare Institute (*Zorginstituut Nederland*; ZiNL) and a subsequent positive decision by the Minister of Health, Welfare and Sports (hereafter referred to as 'the Minister') are a prerequisite for health insurance coverage. ZiNL uses four package criteria for its formal assessment, i.e. effectiveness, cost-effectiveness (i.e. do the effects outweigh the costs), necessity (i.e. do disease severity and the costs per patient justify coverage), and feasibility (i.e. is coverage feasible), with effectiveness being a knockout criterion [14-16].

For interventions that are covered by the basic benefits package, it is assumed that if they do not meet the package criteria anymore (e.g. because of new evidence or because of new alternatives becoming available), they will no longer be delivered by healthcare providers (passive disinvestment), so that explicit exclusion of these interventions (active disinvestment) is not necessary [14]. However, in recent years, the Minister has taken several decisions to exclude specific interventions from the basic benefits package, indicating that the assumption that existing interventions not meeting the package criteria anymore will automatically be (passively) disinvested may not always hold [17, 18]. Hence, active disinvestment may be needed.

Active disinvestment may also be necessary in the case of conditional reimbursement (also called: coverage with evidence development). In the Netherlands, it is possible to conditionally reimburse promising healthcare interventions that do not (yet) meet the package criteria in a special arrangement that is targeted at collecting evidence on the effectiveness and cost-effectiveness of these interventions. In such an arrangement, healthcare interventions will be temporarily reimbursed, provided that additional research will be conducted. After a predetermined time, usually 4 years, the reimbursement of the intervention will be reassessed using the findings from this additional research, which may result in a disinvestment decision [19, 20].

In the international literature, active disinvestment has been described as a very difficult process [21], hampered by contextual aspects such as political barriers [22, 23], a lack of dedicated resources [22], and disinvestment being considered counter-cultural [24]. Furthermore, it has been suggested that, in order to obtain more insight into this difficult process, qualitative studies are needed [23]. Active disinvestment processes in the Netherlands have clearly differed in their outcome. Some attempts at disinvestment resulted in actual disinvestment. Other attempts were terminated at some point, implying unsuccessful disinvestment. Currently, not much is known about active disinvestment processes in the Netherlands and what aspects (package criteria versus other considerations and contextual aspects) determine their outcome. Therefore, here, we aimed to obtain more insight into active disinvestment processes and to explore what aspects determine their outcome. We used a qualitative approach consisting of semi-structured interviews, which enabled us to obtain in-depth insight in the factors influencing the outcome of active disinvestment processes.

Methods

This study focused on policy processes in which the active disinvestment of healthcare interventions or services, which have previously been (conditionally) reimbursed to patients through the basic health insurance, was considered. Active disinvestment includes the full withdrawal, restriction, retraction or substitution of the reimbursement of healthcare interventions or services by means of a policy decision [6, 12]. In policy making, several stages can be distinguished (i.e. the policy cycle): 1) agenda-setting, 2) policy formulation, 3) decision-making, 4) policy implementation and 5) policy evaluation [25]. As the aim of this study was to assess the outcome of active disinvestment processes (i.e. what decision was made with regards to disinvestment), this study mainly focused on the agenda-setting, policy formulation, and decision-making stages of active disinvestment processes.

Theoretical framework

We have used the three essential elements in policy making - actors, ideas and structures [25] - to select distinct cases (see next section), to inform data collection (i.e. to inform the interview guide, see data collection section) and to structure data analysis (i.e. to structure the coding tree, see data analysis section). *Actors* are people or organizations who either 1) try to influence policy to advance their interests or 2) are being influenced by policy. The *ideas* actors have, can range from particular points of view to a widely held, sustained belief system. Arguments are expressions of these ideas. The *structures* in which actors operate are arenas in which policy processes take place and that affect the role that actors and their ideas can play in policymaking [25]. In addition, we have used the model of the organization of the Dutch Healthcare system [26] to guide data collection (i.e. identify categories of actors that may have been involved in the cases studied) and structure data analysis (i.e. to determine actor codes).

Selection of cases

This study focused on five distinct cases to study active disinvestment processes. Possible cases were interventions for which disinvestment was formally and actively considered in the period 2007-2017. This includes interventions that have eventually been actively disinvested, either fully or partially, and interventions for which reimbursement has been maintained. Through screening of policy documents from ZINL and the Ministry of Health, Welfare and Sports (hereafter referred to as ‘the Ministry’) that were found on the websites of ZiNL and the Ministry using specific search terms (e.g. stopping reimbursement, disinvestment), 34 possible cases for which active disinvestment was considered, were identified (see Appendix A). From these 34 possible cases, 5 were selected that were expected to be distinct in 1) the actors, ideas (i.e. reasons for considering disinvestment) or structures involved, and 2) the outcome of the disinvestment process (fully or partially disinvested or reimbursement maintained). Cases were (year of decision in brackets): Benzodiazepines (that are mainly used for anxiety and sleeping disorders; 2007, 2008), Medication for Fabry disease (2012), Quit smoking program (2009, 2011, 2012), Psychoanalytic therapy (2010), and Maternity care assistance (in Dutch: Kraamzorg; 2015). See Appendix B for a description of the selected cases.

Data collection

Semi-structured interviews were conducted with actors who were involved in the disinvestment process for the selected cases. Involved actors were identified from policy documents, through media coverage of the case and through interviews with other actors, and approached through e-mail. New respondents were included for each case until 1) no new information regarding the disinvestment process of the case emerged from the interviews (i.e. informational redundancy/data saturation was reached for the case [27]), 2) all eligible actors for the case were interviewed (i.e. no other actors were involved), or 3) there were no eligible actors left who were willing to participate. The perspectives of the different actor groups were complementary, with the aim to get a complete picture of the disinvestment process of the case. We did not aim to reach saturation with regards to the perspectives of the different actor groups on disinvestment.

The interview topic list was developed for this study and covered actors, ideas, and structures involved in the agenda-setting, policy formulation, and decision-making stage of the disinvestment process. Besides, if relevant, we asked participants to reflect on differences and similarities between the disinvestment process for the case discussed and other reimbursement processes (both investment and disinvestment) they were involved in (if any). The semi-structured interview guide is included in Appendix C. Interviews were audio-recorded and transcribed verbatim.

Ethical considerations

Written informed consent was obtained at the start of the interview. Respondents were explained that only the research team would have access to the audio recordings and transcripts of their interviews, and that they would handle this information confidentially. Furthermore, respondents were explained that, with regards to publications on this study, no names of persons and/or organizations (apart from ZiNL and the Ministry), nor any specific information which would reveal the identity of respondents would be reported. Quotes that were used in this paper were approved by respondents, without them having the possibility to change anything regarding the findings. This research project has been assessed by the Centre for Clinical Expertise (CCE) at the RIVM. The CCE concluded that the research project is exempted from further review by a medical ethics committee as it does not fulfil the specific conditions as stated in the Dutch Medical Research Involving Human Subjects Act (WMO; *Wet Medisch-wetenschappelijk Onderzoek met mensen)*.

Data analysis

Interview transcripts were analysed using thematic content analysis [28]. Transcripts were coded by AR, using a combination of deductive and inductive coding. Text segments from the transcripts were coded using the essential elements in policy making (actors, ideas and structures; see theoretical framework section). The part of the coding tree for the essential element 'ideas' was derived from the Dutch package criteria and the review of Kleinhout-Vliek et al. [14, 29, 30]. The complete coding tree is displayed in Appendix D. Coding was conducted using MAXQDA 2019 [31].

A subset of 15 interviews was also coded by JvdR to assess the reliability of data analysis [32]. This subset was purposively selected to include three interviews on each case and to include interviews with different types of respondents. Within this subset of 15 interviews, there was sufficient consensus between both coders on the text fragments coded and the codes used to code these fragments. Therefore, double coding of the remaining interviews was deemed unnecessary (i.e. remaining interviews were only coded by AR).

To facilitate the analysis, coded text segments were aggregated in three matrices (one for actors, one for ideas and one for structures), with the rows in these matrices displaying the codes and the columns displaying the cases, arranged from most disinvested (i.e. reimbursement fully stopped) to least disinvested (i.e. decision to continue reimbursement). The cells in the matrices contained a short summary of the information in the text segments coded under the respective code for each case. The matrices were inspected by AR, ML and GdW for recurring themes and trends on the disinvestment process itself and aspects determining the outcome of this process. To this end, the authors focused on the differences and commonalities between the studied cases, especially between the cases with similar or very different outcomes. The final main themes were determined through discussion between AR, ML and GdW, focusing on aspects that appeared to have had the largest (or a surprisingly small) impact on the outcome of the disinvestment process in the studied cases. The final themes were checked for plausibility and approved by the second coder based on his/her knowledge of the data from coding a subset of the interviews.

Results

In total, 37 interviews with 37 respondents and an average duration of 53 minutes were conducted between January and December 2018. Of the 37 interviews, 32 were held with one respondent, four with two respondents, and one with four respondents. Of the 37 respondents, 5 were interviewed on multiple cases (counted as separate interviews). Most interviews were administered by one interviewer (AR). In four interviews, a second interviewer joined (GdW or N.J.E. van Vooren) because of the number of respondents taking part in the interviews. A timeline displaying the reimbursement status and the policy reports published on the case was brought to the interviews to stimulate remembrance and to support the conversation (see Appendix E). Table 1 shows the number of interviews that was conducted per case and per respondent group. In four out of five cases, either data saturation was reached and/or all eligible actors were interviewed. In the cases where saturation was reached, no new information on the disinvestment process of the case was obtained from the last interviews conducted. In the quit smoking program case, not all eligible actors were interviewed as we did not succeed to include a politician who was involved.

The remaining part of the results section is structured around the five main aspects influencing the outcome of disinvestment processes, as identified in our analysis. Here, the differences and similarities of these themes between cases are discussed. Appendix F contains a description of these themes for each case, separately. From this point forward, the cases are indicated with an alphabetical code instead of the case name to protect the anonymity of the respondents.

Theme 1: Support for active disinvestment and pressure exerted

In our interviews, respondents described that the degree of support for disinvestment from actors, including the public, was important for the outcome of the disinvestment process. However, the importance of support differed between actors and cases (see Appendix file F and the sections below). Respondents from all cases described that the degree of support for disinvestment was related to actors' views on the case (i.e. health problem and intervention/service concerned), and in some cases to the attention for and framing of the case in the media. Respondents described that for them, as well as for other actors, the degree of support influenced the actions undertaken, and, subsequently, the amount of pressure exerted. From a comparison of the interview data from the studied cases, we found that the pressure exerted had a large effect on the outcome of the disinvestment process: limited pressure was exerted in the cases

Table 1: Number of interviews conducted per case and per respondent group

Case	Respondent group						Total number per case
	Patient representatives[a]	Healthcare providers[b]	Health insurers	National Healthcare Institute (ZINL)	Ministry of health, welfare and sports	Other	
Benzodiazepines	1	2	1	2	2	1[c]	9
Medication for Fabry disease	1	1	1	1	1	1[d]	6
Quit smoking program	1	0[e]	1	1	1	3[f]	7
Psychoanalytic therapy	1	3[g]	1	1	NA[h]	1[c]	7
Maternity care assistance	1	3[i]	1	1	2	0	8
Total over all cases	5	9	5	6	6	6	37

[a] This respondent group includes patients or professionals who acted as patient representatives at the time of the disinvestment process.

[b] The respondent group healthcare providers includes both (leading) individual healthcare providers as well as healthcare provider organizations (e.g. physician associations)

[c] i.e. knowledge institute

[d] i.e. pharmaceutical company

[e] In this case, several interviewees are from interest groups that represent both patients and healthcare providers as well (i.e. different actor groups are united in one organization). Therefore, the healthcare provider perspective has been covered in the other interviews.

[f] i.e. interest groups + advisor of interest groups

[g] In this case, in the first interview with a healthcare provider, we identified two even more involved healthcare providers. Therefore, in total, three interviews with healthcare providers were necessary for this case.

[h] Disinvestment decisions based on (lack of evidence of) effectiveness can be taken by the National Healthcare Institute and do not need involvement by the Ministry. For this reason, the Ministry was not involved in the disinvestment decision regarding psychoanalytic therapy, and was, hence, not interviewed.

[i] Several healthcare provider organizations are involved in maternity care assistance. Therefore, we needed three interviews with healthcare providers to interview all relevant healthcare provider organizations.

that were actually disinvested, while much pressure was exerted in the cases for which reimbursement was maintained. In the following quote, a healthcare provider describes the effect of societal pressure on the outcome of the disinvestment process.

> Healthcare provider, case D, reimbursement maintained:
> *"Yes, I believe that the social pressure, especially all media attention, including emotional arguments, helped a lot to reach this conclusion [not to stop reimbursement] in the end."*

Support from healthcare providers

Respondents described that especially the support from healthcare providers was important for the outcome of the disinvestment process (i.e. has an important effect on the decision that policymakers make):

> Interest group, case C, first disinvested, later reimbursed again:
> *"Respondent: And [the minister] actually had less of a say in that, I think. I think that the social pressure, particularly on this item from the healthcare sector, which she ultimately needs much more, also for the rest of her policy, that it is higher.*
> *Interviewer: In that respect, surely the lobby and the calls from the healthcare sector were very important in this?*
> *Respondent: To reverse it [the decision], yes, definitely."*

By comparing the interview data from the studied cases, we found that the cases where healthcare providers successfully exerted pressure against disinvestment were not disinvested, while cases where healthcare providers did not exert pressure or were unsuccessful in exerting pressure were disinvested. According to respondents, whether healthcare providers were successful in exerting pressure depended on 1) their willingness to exert pressure, subject to a) their level of support for disinvestment of the case, b) whether they feel it fits their role (e.g. whether they feel they have an advisory role which does not comply with exerting pressure) and c) their (financial) interests, and 2) the possibility they had to exert pressure, subject to a) their awareness on how to exert pressure and b) the opportunities provided to them to do so. In the first quote below, a healthcare provider from case B explains that they did not exert pressure because of their belief that disinvestment would improve healthcare delivery (i.e. other healthcare providers would better follow guidelines), suggesting their support for disinvestment. In the second quote a healthcare provider from case A describes his/her difficulty in convincing other actors to join forces (i.e. being successful) in exerting pressure.

> <u>Healthcare provider, case B, partially disinvested:</u>
> *"And, and so in that, up to that point we were like yes, if it actually helps to have it paid for [by patients], [...] and if that helps them [other health care providers] to follow our guideline better, and to give less [of the treatment], yes, then it is fine of course."*
>
> <u>Healthcare provider, case A, fully disinvested:</u>
> *"Interviewer: Was there any party that you managed to get on board?*
> *Respondent: No, not really. [...] I can tell it like it is. At least people paid lip service to this whole story. People understood and supported the arguments. But to say that [name] who was then president of [organization] was like 'will you state once and for all that [case] is indispensable in the Netherlands and at any cost... ', [name] never did that."*

Support from governmental institutions

From our comparison of the interviews, we found that the opportunity provided to actors, including healthcare providers, to exert pressure seemed to depend for a large part on the opportunity actors were given by governmental institutions, who are in charge of disinvestment processes, to be involved in the policy process. We observed large differences between cases in the opportunity respondents were provided to be involved in the policy process, which seemed to depend on the support from governmental institutions for the case at hand. This is illustrated by the quotes below describing the opportunity respondents felt they were provided to be involved in case E, where policymakers described they were not in favour of disinvestment, and case A, where respondents suggested that policymakers were in favour of disinvestment.

> <u>Healthcare provider, case E, reimbursement maintained:</u>
> *"Interviewer: But they [ZiNL] were willing to listen to you in any case?*
> *Respondent: Definitely.*
> *Interviewer: And did they also accept them, um, did they agree with the arguments you put forward?*
> *Respondent: Yes, they did. But I can still remember that they, and it's of course very good that they always wanted to investigate for themselves, were like "okay, we can't accept everything just like that"."*

> <u>Patient organization, case A, fully disinvested:</u>
> *"And well, they [ZiNL] are always going to mention in their advice that they have spoken with – for example, right – in our case [organization name] with so and so. Yes, I can put that into perspective. Those five minutes that it took for us to quickly hand over a box of signatures, well, yes, even if it was a bit longer, that's not really a conversation, is it? There's no way you have really heard us."*

From our comparison of the interview data between cases, we found that the support from governmental institutions also seemed to affect the use of the formal assessment framework in the cases studied. We observed differences between cases in respondents' descriptions of the criteria that were used in the disinvestment process, how these criteria were interpreted, and how these criteria were weighted against one another. For instance, as is illustrated by the quotes below, in case A, where respondents suggested policymakers were in favour of disinvestment, the focus was mainly on whether the treatment was evidence-based, while in case E, where policymakers described they were not in favour of disinvestment, more considerations than strictly related to the four package criteria were taken into account in the policy process.

> <u>Healthcare provider, case A, fully disinvested:</u>
> *"The motive was: we only reimburse things that are evidence-based. Well, a lot of what's in the package just isn't evidence-based so that's kind of a weird reasoning."*

> <u>Policymakers from ZiNL, case E, reimbursement maintained:</u>
> *"You could say it was a very heterogeneous set of effect measures and because of that diversity and heterogeneity, well, it is sort of, it is kind of a multi-factor analysis, right, of all those factors. Are they pointing in the same direction? Do they all contribute to the goal that you want to achieve with [the case]? Well, and that answer was yes in the end."*

Support from patients

Respondents from cases A and B, which were actually disinvested, described that patients in these cases were very vulnerable due to their disease and the societal problems associated with it. Because of this, according to respondents, patients were poorly organized, which made it difficult to get a large group of patients to exert pressure against disinvestment, as is illustrated by the quote below. This lack of pressure exerted may have facilitated disinvestment in these cases.

> Patient organization, case A, fully disinvested:
> *"Yes, for most people it is a taboo, a long-term [treatment] like that. Especially when you're still in the middle of it. So, it was difficult to reach the patients. [...] It was almost impossible, it was a real taboo, there is so much going on when you get such a long-term [treatment]."*

Theme 2: Compassion for current users

Respondents described that the degree of support from actors for disinvestment was affected by whether measures could be taken to ease the effect of disinvestment for patients currently using the intervention at hand. This is illustrated by the quotes from policymakers below in which they describe that they consider disinvestment much more difficult than not to start reimbursement in the first place, especially if disinvestment results in current patients having to stop their treatment or switch to another treatment

> Civil servant from the Ministry, case D, reimbursement maintained:
> *"But you know, you are dealing with people who have been under treatment for years, um, and who are confronted with a new reality overnight. So, that also played a role, also for the Ministry. Like "yeah, you know, what is wisdom, you can't stop people's treatment just like that"."*

> Policymaker ZiNL, case A, fully disinvested:
> *"And we said, "People should be allowed to finish their treatment." That was actually, there was some discussion about that. What were the arguments? I don't know. I think, I don't know how explicit that was but, yes, a reliable government, you started [the treatment] after all, yes."*

Respondents described that if measures could be taken to relieve the effect of disinvestment on current patients, such as stopping reimbursement only for new patients or only restricting reimbursement, they considered disinvestment much more acceptable. This indicates that the possibility to take such mitigating measures impacts the support for disinvestment from actors and, subsequently, the outcome of the disinvestment process.

Theme 3: Role in the health insurance system

From the interviews, we observed that the role appointed to actors by the Dutch health insurance system also affected the outcome of the disinvestment process. In the Dutch health insurance system, two groups of policymakers advise the Minister on reimbursement (policy formulation): ZiNL is involved with reimbursement aspects with regards to content, while policymakers of the Ministry itself both focus at the content and context of reimbursement. Once the Minister decides on reimbursement, health insurers subsequently implement these reimbursement decisions, and patients/healthcare providers are both consulted in reimbursement decisions and in the implementation of these decisions. Every actor can be involved in agenda-setting, although policymakers often have the most distinct role in this phase. In our interviews, respondents described that for the cases studied, actors acted in accordance with the role they have in the system. They described that actors tended to stay away from pursuing actions that they considered to be beyond their formal role. For instance, in the quote below, a health insurer explains that they generally refrained from trying to influence policy formulation and decision-making on reimbursement, as they only consider the implementation of reimbursement decisions to be part of their role.

> Health insurer, case D, reimbursement maintained:
> *"Actually, over the years, we've had less and less of an opinion about this sort of thing. Because we feel, you know, everyone has their part to play in the system."*

Although this finding was most distinct for health insurers, other respondents also regularly discussed their tendency to stick to their formal role. As was described in the interviews, if actors felt that exerting pressure was beyond their formal role, they generally refrained from this. Hence, the formal role of actors affected the pressure that was exerted by these actors in the cases studied and, subsequently, the outcome of disinvestment processes.

Theme 4: Financial interest in disinvestment

From the interviews, we observed that the actions actors took and, subsequently, the outcome of the disinvestment process were also affected by the financial interest of actors in the outcome of the disinvestment process. Respondents described that actors who had a financial interest in maintaining reimbursement were more likely to take action and exert pressure against disinvestment, than actors who did not have a financial interest in maintaining reimbursement, as is demonstrated by the quote below:

> Healthcare provider, case B, partially disinvested:
> *"No, because all those [case intervention] were no longer, there wasn't a patent on them anymore, so they were… It wasn't in those manufacturers interest anymore to interfere. Otherwise it would probably not have been possible [to stop reimbursement]. Then they would have pulled out all the stops, to undo that."*

Theme 5: Role of the formal package criteria

From our interviews, no consistent pattern of the influence of the formal Dutch package criteria (i.e. effectiveness, cost-effectiveness, necessity and feasibility) on the outcome of the disinvestment process was observed. This finding is illustrated by the fact that our study included two cases (i.e. case A and D) that, based on what respondents shared with us during the interviews, seemed to score similarly on the formal package criteria, while having a very different outcome (fully disinvested versus maintained), a different outcome that cannot be explained by the cases scoring differently on any other criterion as well.

Discussion

To our knowledge, this is the first study to describe the agenda-setting, policy formulation and decision-making stages of the active disinvestment process, and to explore aspects influencing its outcome. By using a qualitative approach, we were able to study the reasoning behind the choices made in the active disinvestment process of the five cases studied in-depth and to collect information that has not been included in the formal policy reports written about these cases. Moreover, through interviewing different actors, we were able to look at the disinvestment process from different perspectives, in addition to the formal perspective of policymakers obtained from policy reports.

Our findings indicate that it is more likely for active disinvestment processes to result in a disinvestment decision if 1) there is sufficient support for disinvestment from actors, especially from policymakers and healthcare providers, 2) it is possible to ease the effect of disinvestment for patients currently using the intervention, 3) actors do not have a financial interest in maintaining reimbursement, and 4) actors are not inclined to exert pressure against disinvestment beyond their formal role. Furthermore, we found that the effect of the formal Dutch package criteria on the outcome of the disinvestment process differed between cases.

Comparison with previous studies

Our finding that support, especially from healthcare providers and policymakers, is an important determinant of the outcome of the disinvestment process also has been described by previous studies in which support, especially from clinical leaders, but from the general public as well, was found to be essential for the success of the disinvestment process [6, 33, 34]. Furthermore, a lack of support from decision-makers has been described as a barrier to the implementation of disinvestment [17]. However, it has also been described in previous studies that obtaining support for disinvestment is very difficult as stakeholders often lack the will to support disinvestment because of limited perceived value of disinvestment, which may take some time to realize, and their resistance to immediately “losing” therapy options [12, 35]. Hence, to make actors, especially healthcare providers and policymakers, aware of the value of disinvestment (e.g. quality improvements, reducing displacement) may be essential in order to obtain support and, subsequently, to ensure that a decision to disinvest an intervention is taken and implemented. A review from 2008 focusing on reimbursement decisions for new medicines also found healthcare providers and administrators (i.e. policymakers) to be most influential in reimbursement decisions [36], indicating that this aspect may be similar in investment and in disinvestment decisions.

In our study, we found that limiting the effect of disinvestment on current patients increased support for disinvestment and, subsequently, facilitated actual disinvestment, as actors were reluctant to disrupt treatment of current patients. This has also been found in two previous studies in which citizens and policymakers expressed a reluctance to disrupt treatments because of the negative effect this may have on current patients, e.g. anxiety because of losing the current treatment option and side-effects of stopping/changing treatments [6, 37]. Hence, when considering the disinvestment of healthcare interventions, it is important to consider if and how the treatment of current patients could be (partially) continued. This consideration may be less relevant in the context of investment as, in the investment context, patients are not yet receiving the appraised intervention, and hence, no treatment will be disrupted if a decision not to reimburse the intervention is taken. This indicates that different considerations may be relevant in the context of disinvestment compared to the context of investment.

In this study, we found that actors’ financial interests influence the outcome of the active disinvestment process through the action’s actors undertake. Similar findings have been described by previous studies on the disinvestment/de-implementation of low value care. For instance, van Egmond et al. found that financial incentives influence the de-implementation of low-value care by healthcare providers [38]. Furthermore, Haas et al. and Robinson et al. describe that there is a lack of financial incentives for healthcare providers to engage in disinvestment [24, 39]. Further research is needed to determine how changing financial incentives may facilitate disinvestment. To our knowledge, our finding that actors’ inclination to exert pressure because of the role appointed to them by the health insurance system influences the outcome of active disinvestment processes has not been found by previous studies on the disinvestment of healthcare interventions.

In the Netherlands, a formal assessment framework is in place for the appraisal of new healthcare interventions for reimbursement. However, in this study we found that the formal criteria from this assessment framework for new additions to the basic benefits package (i.e. effectiveness, cost-effectiveness, necessity and feasibility) have been inconsistently applied in disinvestment decisions. This finding may be problematic as a structured, evidence-based process has been described in a review from 2015 as a facilitator for (the implementation of) disinvestment [17]. Furthermore, consistency in the rationale underlying decisions has been described as an important requirement for decision-making to be considered fair [40]. Therefore, it is important for policymakers to ensure that the criteria underlying disinvestment decisions are consistently applied. Such a consistent assessment framework may also facilitate the selection of the potentially most successful (i.e. most likely to result in actual disinvestment) cases for active disinvestment processes.

Previous studies have described that, although broad stakeholder engagement is considered essential for the success of disinvestment, it is considered very difficult to sufficiently organize this [17, 22, 24]. Although this aspect has not been specifically addressed in the current study, we did find that the possibility for stakeholders to be involved in the disinvestment process differed between cases, depending on the support from policymakers for disinvestment. Furthermore, we found that the engagement of patients was difficult, especially in cases where the patient group was in a vulnerable situation and, because of this, poorly organized.

Strengths and limitations

In this study, we have used a qualitative approach which enabled us to obtain in-depth insight into the factors that influence disinvestment processes and their outcomes. Furthermore, by interviewing many different actors who have perceived the disinvestment process from different perspectives, we were able to obtain a comprehensive view of the disinvestment processes studied. The perspectives of all the different actors were of added value and treated equally, complementing each other in the reconstruction of the disinvestment processes. Moreover, as a sample (40%) of the interview transcripts was double coded, and the matrices were checked for recurring themes by three researchers, we ensured the reliability of data analysis [32]. In addition, as the disinvestment process of the studied cases took place several years ago, it may have been perceived as a closed chapter by respondents, making elaborating on the cases less sensitive and reflecting on it easier.

However, our study also has some limitations. Firstly, we have selected 5 cases from a list of 34 identified cases. Although we aimed to select 5 distinctive cases from this list, we may have missed aspects determining the outcome of the disinvestment process that may have been more pronounced in the non-selected cases. Furthermore, as we identified the 34 possible cases from policy documents, we may have introduced bias, as we could only select cases that made it to the stage of a policy document. Because of this, we may have missed cases for which the disinvestment process was terminated at a very early stage, before any policy document was written. However, even though it is clear that some information may have been omitted due to case selection, we did include a distinct selection of cases to maximize the transferability of our findings to other cases.

Secondly, although we tried our best to have all the relevant topics discussed during the interviews, it may be that respondents (intentionally or unintentionally) left some relevant information out. Especially one of the five cases studied (i.e. case D) still appeared to be sensitive to respondents, indicating that it may be more prone to information omission. We tried to overcome this limitation by including multiple respondents for each case. No conflicting information was obtained from the interviews with the different respondents, indicating that there are no clear signs of information omission. Furthermore, the older cases may have also been more prone to information omission because respondents may have had trouble remembering the relevant information. We tried to overcome this by showing respondents a timeline displaying policy reports and the reimbursement status to stimulate remembrance. In the interviews, this timeline appeared to be helpful for respondents in remembering the disinvestment process.

Thirdly, in the quit smoking program case we were not able to interview all eligible actors because we did not succeed in including a politician who was involved in this case. As politicians played a major role in this case, it would have been informative to interview a politician. However, as the political process was extensively discussed in the remaining interviews on the case, we expect the information omission by not including politicians to be limited.

Finally, this study has been conducted in a Dutch setting. As disinvestment processes are often context-specific [21], one should take the differences between healthcare systems and other contextual factors into account when considering the implications of this study for other healthcare systems. For instance, in other healthcare systems, the roles may be differently divided between the different actors than is the case in the Dutch healthcare system. Moreover, actors may have a different tendency to stick to these roles. This may affect the importance of this aspect for the outcome of disinvestment processes in other healthcare systems. Therefore, to find out which aspects determine the outcome of disinvestment processes in other contexts, future research may consider conducting a similar assessment as has been conducted in this study. Note, however, that conducting multiple interviews per case is dependent on the willingness of key actors to share their experience. In our current study, almost all approached actors were willing to participate. We believe that this large willingness to participate may have been caused by the fact that the disinvestment processes of the cases took place several years ago. Therefore, to increase the willingness of key actors to participate in future studies, we recommend to select cases that may be perceived as a closed chapter, but that are recent enough for actors to sufficiently remember the disinvestment process.

Implications

The results of this study may have implications for future active disinvestment processes. If policymakers are interested in selecting candidate interventions for which the disinvestment process is more likely to result in a disinvestment decision, they are recommended to select interventions for which there is support for disinvestment from actors, especially from healthcare providers. Furthermore, interventions for which support can be easily obtained, for instance because of the possibility to ease the consequences of disinvestment for current patients, are suitable candidate interventions as well. Moreover, also interventions for which actors do not have (strong) financial interests in maintaining reimbursement are suitable candidates for disinvestment.

After having selected candidate interventions for disinvestment, policymakers are recommended to increase support for disinvestment to facilitate the disinvestment process. As support from healthcare providers appeared to be important, policymakers are recommended to start with increasing support among this actor group. To this end, healthcare providers who are not aware of all the potential merits of disinvestment should be made aware of these merits in addition to the possible disadvantages they may already be aware of (i.e. losing therapy options for new patients and disrupting treatment of current patients). In order to make these healthcare providers aware of the merits, the scientific literature suggests that the emphasis of disinvestment processes should be on safety, and quality improvements such as avoiding side effects of ineffective care, instead of cost savings as this makes disinvestment more acceptable to healthcare providers [18, 41]. Furthermore, it is important to make these healthcare providers aware of the opportunity costs of providing the intervention that will be disinvested. For instance, healthcare providers should be explained that, by disinvesting the intervention, their time and efforts can be devoted to interventions that generate more health outcomes. Moreover, healthcare providers should be made aware of the displacement taking place in the healthcare system because of the reimbursement and usage of interventions that are unsafe or not (cost-)effective. In addition, as easing the consequences of disinvestment on current patients appeared to increase support, it is recommended to explore the opportunities to do so for candidate interventions for disinvestment. In order to facilitate disinvestment, policymakers are also recommended to explore the financial interests that actors have with regards to disinvesting or maintaining reimbursement of the intervention and the mechanisms underlying these financial interests. By being aware of this, policymakers can better anticipate on the pressure that will be exerted by actors because of these financial interests.

It is important for policymakers to be aware of the fact that different considerations may be relevant in the disinvestment context compared to the investment context. For instance, the reluctance to disrupt treatment for current patients may be highly relevant with regards to disinvestment decisions, while not being relevant in the investment context. As consistency and transparency in the rationale underlying disinvestment decisions is an important requirement for the policy process to be considered fair [40], we recommend policymakers to develop, consistently apply and transparently communicate on an assessment framework for disinvestment decisions, taking into account considerations that are specifically relevant in the disinvestment context.

Implications for conditional reimbursement

Conditional reimbursement, or coverage with evidence development is the reimbursement of (new) healthcare interventions under a specific condition, often the requirement of collecting real-world (cost-)effectiveness data. It has been proposed as a policy-tool to delay the final reimbursement decision, decreasing the uncertainty on the (cost-)effectiveness and/or budget-impact of new interventions, while making these interventions available to patients at an early stage [42, 43]. However, conditional reimbursement has also been described as creating a wedge effect, i.e. giving suppliers a “foot in the door” while creating challenges for decision-makers when the reversal of (conditional) reimbursement is warranted [12]. As the intervention has already been provided to patients, reversal of reimbursement implies the withdrawal of treatment, which has been described in the scientific literature as far less acceptable to actors than withholding treatment [42, 44]. However, the specific nature of conditional reimbursement processes, provides policymakers with opportunities to address the aspects influencing the likelihood of an active disinvestment process to result in a disinvestment decision, as identified in our study. For instance, conditional reimbursement has the advantage that support for disinvestment can be ensured before starting reimbursement. Therefore, to ensure support, policymakers are recommended to make agreements with actors, before starting reimbursement, on the criteria the intervention should meet to maintain reimbursement after the evaluation period. Furthermore, we recommend policymakers to change the default outcome of conditional reimbursement from ‘reimbursement is maintained if the intervention meets the criteria’ to ‘reimbursement is reversed, unless the intervention meets the criteria’. Although this difference may seem trivial, it has shown to be important psychologically [45, 46]. Subsequently, policymakers are recommended to make sure that actors understand and agree that if the intervention does not meet the agreed upon criteria at the end of the reimbursement period, it will be disinvested. In this way, policymakers ensure that actors know that care is only provided under the precondition that it can be withdrawn later on, which makes withdrawing treatment similar to withholding treatment from a psychological point of view [42, 44].

Conclusions

In conclusion, this study indicates that contextual aspects such as support, institutional structures and financial interests are important determinants of the outcome of active disinvestment processes. Furthermore, the possibility to relieve the consequences of disinvestment for current patients appeared to be important to increase the acceptability of disinvestment. These results provide researchers and policymakers with insight into disinvestment processes in the Netherlands. Furthermore, the results can be used by policymakers in the selection of future candidate interventions for disinvestment and in future disinvestment processes to increase the likelihood of the disinvestment process to result in an actual disinvestment decision. Moreover, the results can be used by policymakers to prevent some of the challenges inherent in reversing reimbursement after a period of conditional reimbursement.

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Appendices

Appendix A: Identified cases, categorized (selected cases are marked green)

Type of disinvestment	**Main reason for disinvestment (according to policy documents):**			
	Effectiveness	**Cost-effectiveness / budget cuts**	**Necessity**	**Feasibility**
Full disinvestment	NESS handmaster (2007)	🟩 Quit smoking interventions (2012)		Diane-35 pill (2014)
	Allergy-free covers (2009)	🟩 Medication Fabry disease (2013)	Stand-up-chair (2009)	Non-complicated extractions (2011)
	Acetylcysteine (2010)	Medication Pompe disease (2013)	Rollator, crutches, walker (2013)	Circumcision (2013)
	🟩 Psychoanalytic therapy (2010)	Ranibizumab (2015)	Contraceptives (2011)	
	Intravesical sodium chondroitin sulphate/ hyaluronic acid (2014)	Statins (2009)	Helmet therapy (2013)	
	Renal denervation (2017)		Second opinion (2016)	
	Radiofrequent denervation (2016)		Fax machines for auditory impaired (2009)	
	Contralum ultra (2016)		🟩 Maternity care assistance (2016)	
	Paracetamol-codeine (2013)		Intracavernous fentolamine /papaverine (2009)	
Restriction		Antacida (2012)	Dentist (2011)	🟩 Benzodiazepines (2009)
		Diet advise (2012)		
		Fertility treatment (2013)		
		Anti-depressants (2011)		
		Physiotherapy (2011 & 2012)		
Retraction			Incontinence products (2012)	
		Curative mental healthcare (2012)		
Replacement				

Appendix B: Description of the selected cases

Benzodiazepines

Benzodiazepines are sedative medicines that are mainly used for sleep and anxiety disorders. The policy process for the disinvestment decision took place in 2007 and 2008. Starting January 2009, benzodiazepines are no longer reimbursed through the basic benefit package. Exceptions were, however, made for five indications: epilepsy, therapy-resistant anxiety disorder, multiple psychiatric problems, palliative sedation, and therapy-resistant muscle spasms because of neurological disorders (the latter concerns only diazepam, no other benzodiazepines).

Medication for Fabry disease

Fabry disease is a hereditary, chronic and slowly progressive disease. In early disease stages, patients mainly suffer from pain in hands and feet, and a high body temperature. In later stages, patients experience symptoms of kidney failure, heart disorder and stroke. The disinvestment process concerned two orphan medicines, Agalsidase alpha and Agalsidase beta. The aim of these medicines is to decrease disease symptoms and to slow the progression of the disease. The medicines have been conditionally reimbursed, and were reassessed in 2012. In the reassessment, it was decided to keep reimbursing the medicines after (successful) price negotiations.

Quit smoking programme

The quit smoking programme includes two treatment options: 1) behavioural counselling, and 2) behavioural counselling combined with medicines. Starting January 2011, the quit smoking programme was included in the basic benefit package. A year later, starting January 2012, the quit smoking programme was disinvested from the basic benefit package. One year after that, starting January 2013, the quit smoking programme was, again, included in the basic benefit package and has remained reimbursed since. For our study, we mainly focused on the disinvestment in 2012 and the re-introduction in 2013.

Psychoanalytic therapy

The reimbursement decision, actually, concerned two therapies within the psychoanalysis field: psychoanalytic therapy and long-term psychoanalytic psychotherapy (LPPT). Psychoanalytic therapy is mainly focused on opening up subconscious aspects of dysfunctional behaviour and dysfunctional experiences. It consists of four to five consultations a week, for five to ten years, in which the patient is laying down on a sofa. The psychoanalyst is sitting out of sight and mainly listens and reacts to what patients tell him/her. LPPT is a less intensive form of psychoanalytic therapy. It consists of one or two consultations a week, for one to two years. Instead of lying on the sofa with the analyst out of sight, the patient is sitting down, facing the analyst. The disinvestment process mainly took place in 2009 and 2010. Starting 2010, psychoanalytic therapy is no longer reimbursed through the basic benefit package. LPPT was not disinvested.

Maternity care assistance

Maternity care assistance (in Dutch: kraamzorg) is postpartum care for new-borns and their mothers. The maternity care assistant provides care at home[1] in the first days (usually 8 days) after birth, under the supervision of a midwife. The main aim of maternity care assistance is to monitor and support a quick recovery of the mother and the appropriate development of the new-born. The main tasks of the maternity care assistant are nursing care for the new-born and the mother, providing health-education, signalling health problems, (breast) feeding support, and support with basic household tasks (to promote a hygienic environment). In 2012, in a Dutch tv-show, the minister of Health, Welfare and Sports called the public for suggestions to save healthcare costs. Some reactions to this call concerned stopping the reimbursement of maternity care assistance. Therefore, the minister asked the National Healthcare Institute (ZINL) for an advice regarding the reimbursement of maternity care assistance. In 2015, ZINL advised the minister to keep reimbursing maternity care assistance and this advice was followed by the minister.

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[1] In the Netherlands, many women with low-risk pregnancies give birth at home. Women that do give birth at the hospital or a birth center are often discharged after a few hours to recover at home as well. Only women/newborns with severe complications are admitted to the hospital for a longer time.

Appendix C: Semi-structured interview guide

Introduction

As was described in the information letter, this study of the RIVM concerns disinvestment. Disinvestment is 1) the complete withdrawal of reimbursement, 2) the restriction of reimbursement, 3) the retraction of budget from reimbursement or 4) the replacement of healthcare interventions by alternatives.

In this study, the focus is on the disinvestment of healthcare interventions from the basic benefit package of health insurance. It concerns care which has been reimbursed to patients before. (Hence, it also concerns care which has erroneously been delivered at the expense of the basic benefit package because a lack of effectiveness had not yet been determined.)

To obtain more insight into active disinvestment processes and to explore what aspects determine their outcome, we interview stakeholders who were involved in disinvestment processes. In these interviews, we focus on a number of cases of disinvestment. These are both cases in which a disinvestment decision has been taken, as well as cases that only have gone through some stages in the disinvestment process, because of which no disinvestment decision has been taken. For instance, this concerns cases on which only an advice of the National Healthcare Institute has been written.

This interview concerns [case]. We approached you for an interview as you have been involved in the disinvestment process of this case.

To be able to analyse the interviews in an objective way, I ask you for your permission to record the interviews using an audio recorder. This recording will be used to write down (transcribe) the interview for analysis. We will send you the transcribed interview, so that you have the opportunity to check it for inaccuracies. We will handle both the audio recording as the transcribed interview confidentially. Only the research team will have access to this information.

The results of this study will be published. We may use one of your quotes in this publication to substantiate the results. We will, however, ensure that both the results and the quotes will not contain information that may reveal your identity.

Is everything clear? Do you agree with:
- Participation in this study?
- The audio recording and writing down of the interview?
- The inclusion of the results and possible quotes in a publication?

Do you have any questions before we will start with the interview?

[ask respondent to fill-out and sign informed consent form, start audio recording]

Interview

Introduction

Topic	**Questions**
General	As discussed before, this interview concerns [case].
	Later on, I will pursue this case further. However, as a starting question: could you give a short summary of the case and your involvement in it?

Agenda-setting

Topic	**Possible questions**[2]
General	What was the reason to consider whether the reimbursement of [case] should be changed?
Actors	Which parties/stakeholders were involved?
Ideas	What was your idea/opinion on the case?
	Which ideas/opinions did other parties/stakeholders have on the case?
	Which arguments were raised?
Structures	What was your role in this case? Formally? In practice?
	What role did other stakeholder have? Formally? In practice?
	To what extent did you have an influence on whether reimbursement would be changed?
	To what extent did other stakeholders have an influence on whether reimbursement would be changed?
	What was the role of the different arguments?
	To what extent were the arguments a decisive factor in the disinvestment decision?

Policy formulation

Topic	**Possible questions**
General	Which options for reimbursement have been discussed?
	Have other policy options been discussed in which reimbursement would not be changed?
	How were these policy options identified?
	How were these policy options assessed?
Actors	Which parties/stakeholders were involved in the identification and the assessment of the different policy options?

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[2] The exact questions that were asked were formulated during the interviews, based on the responses of the respondent. However, interviewers made sure that all topics were covered during the interviews.

Ideas	Which ideas/opinions did these parties/stakeholders have on the case?
	What was your idea/opinion on the different policy options?
	Which ideas/opinions did other parties/stakeholders have on the policy options?
	What arguments were raised by the parties/stakeholders?
Structures	What was your role? Formally? In practice?
	What role did other stakeholder have? Formally? In practice?
	To what extent did you have an influence on the identification and assessment of policy options?
	To what extent did other parties/stakeholders have an influence on the identification and assessment of policy options?
	What was the role of the different arguments?
	To what extent were the arguments a decisive factor in the identification and assessment of policy options?

Decision-making

Topic	**Possible questions**
General	What was the final decision?
	How has this decision been taken?
Actors	Which parties/stakeholders were involved?
Ideas	Which ideas/opinions did these parties/stakeholders have on the case?
	What arguments were raised by the parties/stakeholders?
Structures	What was your role? Formally? In practice?
	What role did other stakeholder have? Formally? In practice?
	To what extent did you have an influence on the final decision?
	To what extent did other parties/stakeholders have an influence on the final decision?
	What was the role of the different arguments?
	To what extent were the arguments a decisive factor for the final decision?

Actors who were involved, but have not yet been discussed/actors who were not involved

Topic	**Possible questions**
Involved, but not yet discussed	Were there any other parties/stakeholders that were involved in the disinvestment process on the case, but have not yet been discussed?
	What were their ideas/arguments raised by them?
	What was their role in the process?
Not involved	Were there any parties/stakeholders who were not involved in the disinvestment process on this case, but who are affected by the outcome of this process?
	What were their ideas/arguments raised by them?
	Why were they not involved in the process?

Comparison with other cases (if applicable)

Topic	**Possible questions**
Disinvestment cases	From your perspective, did the disinvestment process of [the case discussed] differ from disinvestment processes of other cases? - Comparable cases? - Cases in which a decision was/was not taken?
Investment decisions	From your perspective, did the disinvestment process of [the case discussed] differ from the assessment of new treatments for the inclusion in the basic benefit package? - Actors - Ideas - Structures

Closing

- Are there other things you would like to discuss on this case? Are there any relevant issues that have not yet been discussed?
- Do you have any written/internal documents on this case I may have a glance over?
- Are there other things you would like to discuss on disinvestment processes?
- Are there other stakeholders you think I should interview on this case? If so, who could we approach for this?
- Thank respondent and close off

Appendix D: coding tree

Conceptual framework	**Main codes**	**Subcodes**		
Essential elements	Actors	Ministry of Health, Welfare and Sports		
		Dutch Healthcare Institute (ZiNL)		
		Patient(s) (organizations)		
		Health care provider (organizations)		
		Manufacturers		
		Health insurers		
		Media		
		Knowledge institutes		
		Politicians/ members of parliament[1]		
		Minister/ secretary of state of Health, Welfare and Sports		
		Health Council		
		ZonMW (The Netherlands Organisation for Health Research and Development)		
		SER (Social and economic council)		
		Health funds		
		Partnership quit smoking		
		Stivoro (organisation against smoking)		
		Tobacco industry/lobby		
		Other		
	Ideas	Effectiveness	Common practice	
			Efficacy	
			Effectiveness	
			Variability	
			Strength of evidence	*Level of evidence*
				Amount of research
				No evidence is not evidence of ineffectiveness
				Methodological quality
			Statistical significance	
			Clinical relevance	

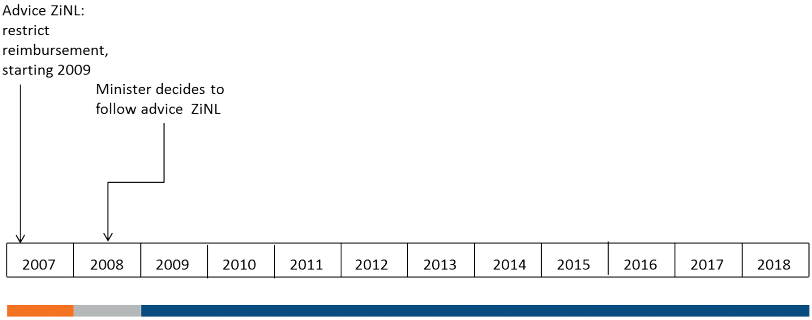
Conceptual framework	**Main codes**	**Subcodes**		
			Patient relevance	
			Safety/adverse events	
			Limitations in doing research	
			Necessity for further research	
			Mechanism of action	
		Cost-effectiveness	ICER vs reference value	
			Costs (per unit of the intervention)	
			QOL/QALYs	
			Uncertainty	
			Strength of evidence	Methodological quality
			Necessity of further research	
		Necessity	Necessity to insure	Definition of illness
				Definition of treatment/ what is covered by the basic benefit package
				Individual Cost
				Individual responsibility
				Moral Hazard
				Range of Normality
				Societal responsibility
			Medical Necessity	Medical Necessity
				Morbidity/Severity
				Need
				Rule of Rescue
			Other necessity aspects	Dignity
				Equity/fairness/justice
				Human Right
				(No) Alternative
				Patient-diagnosis
				Similar Treatments
				Number of Patients
				Societal impact
				Societal functioning
				Vulnerability/compassion
				Has been used to treat patients/reimbursed before
		Feasibility	Support	
			Organization of care	

Conceptual framework	**Main codes**	**Subcodes**		
			Indication and administration	
			Financing	
			Legal and ethical	
			Consultation and anticipation	
			Commencing date	
			Consequences for healthcare consumption	*Substitution*
			Budget impact	Societal
				Health care budget
		Other	*Considered/fair process*	*Agenda-setting (sitting duck, arbitrariness)*
				Policy-development (criteria/ process)
				Decision-making
				Implementation
			Healthcare delivery	*Tailored care*
				Quality of care
			Undesirable use	
			Autonomy healthcare providers	
			Trustworthy government	
			Disappearance/ erodation of a field	
			Reimbursement status gives a signal	
			Values	*Solidarity*
				Accessibility
			Prevention (value of)	*Signaling health problems*
			Emotional arguments	*You want to be able to do something for your patient as a temporary relieve*
			Financial arguments	*Need to cut healthcare budget*
				Profit/returns
				Having budget/financing
				Sunk costs
				Defend price setting

Conceptual framework	**Main codes**	**Subcodes**		
	Structures	Social/cultural	*Societal vision/ image/ idea*	
			Clinical vision/ image/ idea	
		Economic		
		Political	*Feeling of urgency*	
			Momentum	
			Compromise	
			Societal pressure	
			Political pressure	
			Loss of face/ reputation	
			Political vision	
			Polarised debate	
		Institutional/ regulatory	*Roles/ tasks organizations*	
			Drug regulations/ approval	
			Withdrawing government	
		Historical		
		Financial		
		Other policies		
		Other reimbursement decisions		
		Investment vs disinvestment		
		Other reports		
		Other		
Policy cycle	Agenda -setting			
	Policy development	*Reimbursement options*		
	Decision-making			
	Implementation	*Side effects process*		
		Uncertainty about reimbursement		

[1]Codes in italics have been inductively added to the coding tree during the coding process

Appendix E: Timelines brought to the interviews



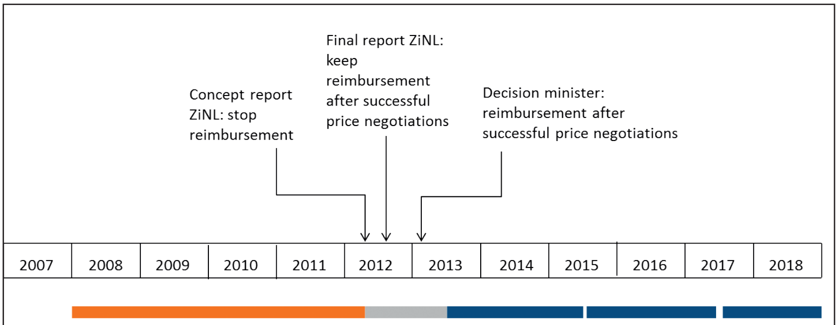
🟧 = Reimbursement

⬜ = Transition year, benzodiazepines still reimbursed, aiming to decrease chronic use

🟦 = Benzodiazepines only reimbursed for

1) Epilepsy
2) Therapy-resistant anxiety disorder (no reaction to ≥2 antidepressants)
3) Multiple psychiatric disorders
4) Palliative sedation

Benzodiazepines

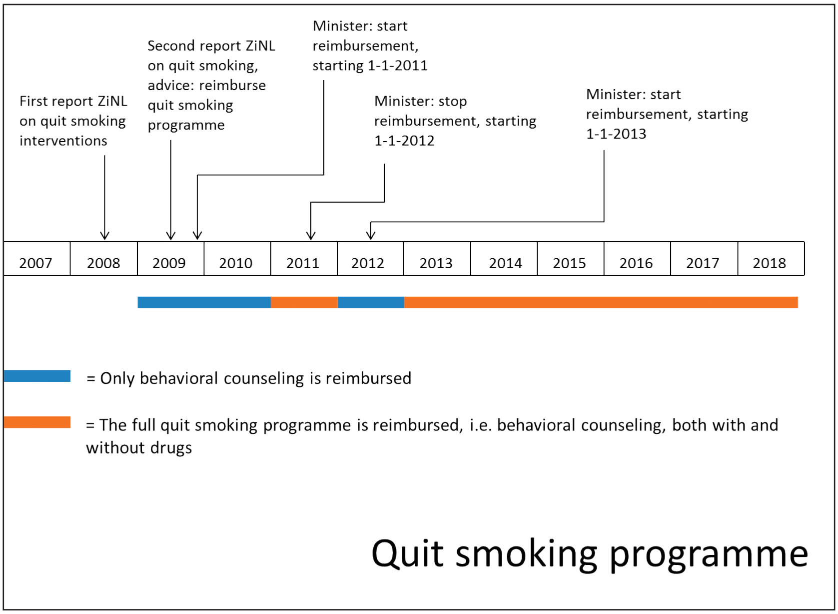


🟧 = Conditional reimbursement

⬜ = Uncertainty regarding reimbursement

🟦 = Reimbursement after price negotiations with pharmaceutical companies

Medication for Fabry disease



First report ZiNL on quit smoking interventions

Second report ZiNL on quit smoking, advice: reimburse quit smoking programme

Minister: start reimbursement, starting 1-1-2011

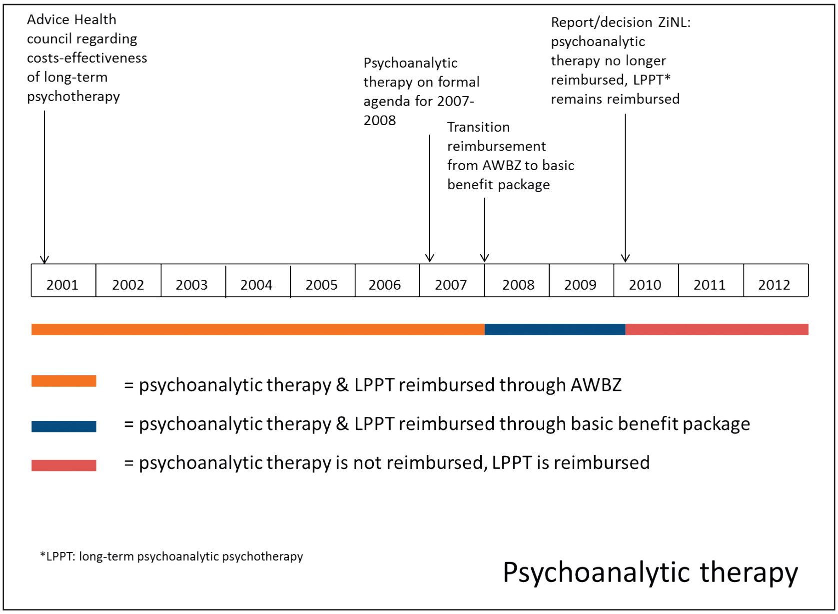
Minister: stop reimbursement, starting 1-1-2012

Minister: start reimbursement, starting 1-1-2013

🟦 = Only behavioral counseling is reimbursed

🟧 = The full quit smoking programme is reimbursed, i.e. behavioral counseling, both with and without drugs

Quit smoking programme



Advice Health council regarding costs-effectiveness of long-term psychotherapy

Psychoanalytic therapy on formal agenda for 2007-2008

Transition reimbursement from AWBZ to basic benefit package

Report/decision ZiNL: psychoanalytic therapy no longer reimbursed, LPPT* remains reimbursed

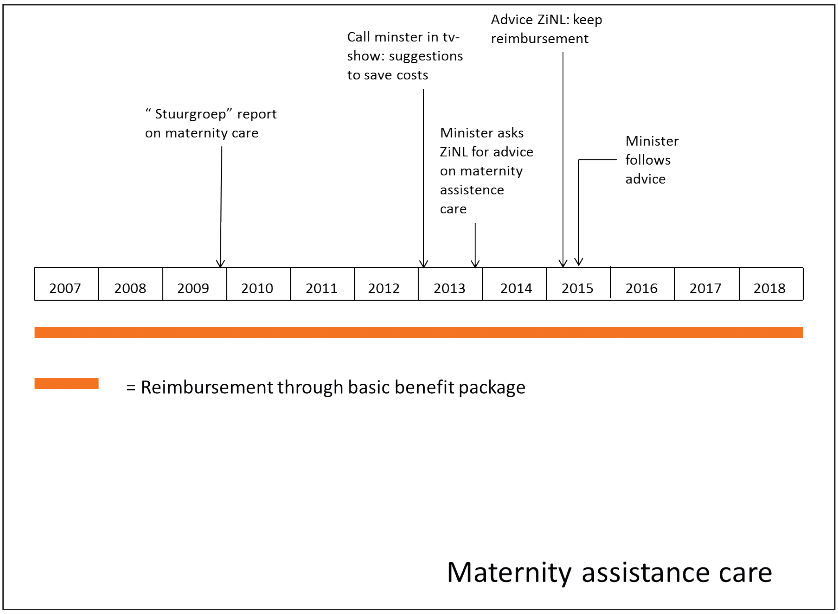
🟧 = psychoanalytic therapy & LPPT reimbursed through AWBZ

🟦 = psychoanalytic therapy & LPPT reimbursed through basic benefit package

🟥 = psychoanalytic therapy is not reimbursed, LPPT is reimbursed

*LPPT: long-term psychoanalytic psychotherapy

Psychoanalytic therapy


Call minster in tv-show: suggestions to save costs
Advice ZiNL: keep reimbursement
" Stuurgroep" report on maternity care
Minister asks ZiNL for advice on maternity assistence care
Minister follows advice
2007
2008
2009
2010
2011
2012
2013
2014
2015
2016
2017
2018
= Reimbursement through basic benefit package
Maternity assistance care

Appendix F: Themes at case level

Themes	Cases	
	A	B
	Fully disinvested	Partially disinvested
Support for active disinvestment and pressure exerted - Healthcare providers - Governmental institutions - Patients	Respondents described that there was much societal support for disinvestment, because of the negative view of society on the case. Healthcare providers and patients were against disinvestment but were not sufficiently able to exert pressure. Respondents described that patients were very vulnerable and, consequently, poorly organized. Furthermore, respondents described that healthcare providers were difficult to mobilize because the small group of healthcare providers was highly divided. According to respondents, governmental institutions were not very open to the input of healthcare providers and patients in this case.	Respondents described that there was broad support for disinvestment among actors, especially because disinvestment was only partial (i.e. several patient groups were excluded from disinvestment) and because of the negative connotation of the case. They described that, although healthcare providers preferred a more passive approach to disinvestment, they generally supported disinvestment. Respondents described that patients were vulnerable and, consequently, poorly organized. Furthermore, the use of the case intervention is not restricted to a certain patient group. Respondents described that, for these reasons, patients only had a minor role.

C	**D**	**E**
First disinvested, later reimbursed again	**Reimbursement maintained**	**Reimbursement maintained**
Respondents described that political and societal support was very important in this case. Throughout the years, the vision on the case shifted, which affected support for disinvestment. Furthermore, they described that healthcare providers and interest groups had a large role in reversing disinvestment, because policy makers needed the support from them for other policies as well. Respondents described that, initially, healthcare providers did not exert much pressure against disinvestment. However, after disinvestment took place, healthcare providers exerted much pressure to reverse disinvestment, among others because of a shifted vision on the case. Respondents described that patients did not have a role in this case because they felt that it would not be considered societally acceptable for them to take any action.	Respondents described that there was broad support for reimbursement, which was driven by the framing of the case in the media. This broad support was essential in maintaining reimbursement. Respondents described that healthcare providers were very successful in exerting pressure in this case. They were in constant interaction with ZiNL, who carefully considered their input. Furthermore, respondents described that healthcare providers helped patients to exert pressure. In earlier phases of the disinvestment process, patients had a limited role. But later on, when there was media attention, patients could play a somewhat larger role.	Respondents described that some citizens and civil servants were against reimbursement of this case. These people contributed to agenda-setting. However, according to respondents, as other actors were involved in the later stages of the disinvestment process, there was broad support for maintaining reimbursement in these stages. They described that this contributed to reimbursement being maintained. Respondents described that healthcare providers cooperated with each other in exerting pressure. Furthermore, ZiNL was very much open to the input of healthcare providers, with the aim of knowledge sharing. Respondents suggested that these aspects may have facilitated reimbursement being maintained. According to respondents, patient groups were not very visible in the disinvestment process.

Themes	**Cases**	
	A	**B**
	Fully disinvested	**Partially disinvested**
Compassion for current users	In this case, it was decided to stop reimbursement only for new patients. Current patients could finish their treatment. Respondents described that this was important because actors were reluctant to disrupt treatment. Therefore, they implied that the possibility to stop reimbursement only for new patients substantially contributed to the support for disinvestment.	In this case, several exceptions to disinvestment were made. Respondents described that actors felt that patient groups that really benefited from the case intervention, could still receive reimbursement for this intervention. This facilitated the support for disinvestment for the patient groups who benefitted to a lesser extent from the case intervention.
Role in health insurance system	According to respondents, as the disinvestment decision was only substantiated by the lack of evidence of effectiveness of the case intervention, the role of the Ministry was limited because they consider decisions based on just effectiveness to be beyond their role (i.e. ZiNL is in charge when it only concerns effectiveness).	Respondents described that healthcare providers did not consider it to be their role to exert pressure. For this reason, they tended to stick to comments with regards medical aspects and stay away from exerting pressure. Furthermore, respondents described that health insurers only considered implementation (of disinvestment decisions) as part of their role. For this reason, they were not involved in policy development and decision-making. Moreover, the ministry considered policy development beyond their role. Therefore, they tried to refrain from involvement in policy-development.

C	D	E
First disinvested, later reimbursed again	**Reimbursement maintained**	**Reimbursement maintained**
NA[1]	There was much variation in effectiveness between patient groups. However, respondents described that, in this case, it was not possible to discern patients who would benefit from the case intervention from patients who would not benefit from this intervention, implying that no exceptions could be made. Respondents described that actors did not want to stop reimbursement for all patients because this would disadvantage patients who could benefit from the case intervention. This contributed to disinvestment being maintained for all patients.	It was not possible to discern patient groups who were more in need for the case intervention from patient groups that were less in need for this intervention, implying that no exceptions could be made. Respondents described that actors were reluctant to stop reimbursement for all patients because this would disadvantage patients who were in greatest need for the case intervention. According to respondent, this contributed to reimbursement being maintained for all patients.
Respondents described that although health insurers supported disinvestment, they did not consider involvement in reimbursement decisions to be their role. Furthermore, ZiNL also played a limited role, according to respondents, as the decision was mainly political, which ZiNL considers to be the role of the Minister/Ministry.	Respondents described that health insurers felt that involvement in reimbursement decisions does not fit their role. Respondents suggested that this may explain the lack of action undertaken by health insurers. Furthermore, respondents described that the Ministry and ZiNL felt that their role was sequential. First, ZiNL was in the lead. Subsequently, the Ministry/Minister was in the lead. Respondents described that the governmental institutions tended to stick to these roles.	Health insurers described that, although they normally would abstain from being involved in disinvestment processes, in this case, they did exert some pressure because disinvestment would affect provider payment, which they do consider to be part of their role.

Themes	Cases	
	A	B
	Fully disinvested	**Partially disinvested**
Financial interest in disinvestment	NA[1]	Respondents described that because the case intervention was no longer patented, manufacturers did not exert any pressure against disinvestment.
Role of formal package criteria:		
- Effectiveness	According to respondents, lack of evidence of effectiveness from comparative studies was the main reason for disinvestment. Patient experiences of effectiveness did not play a role.	According to respondents, lack of effectiveness, combined with many side-effects was the main reason for disinvestment. Respondents described that effectiveness was also the main reason to make exceptions to disinvestment.
- Cost-effectiveness	Respondents described that the high costs of the case intervention per patient was important. Apart from this, cost-effectiveness did not play a role.	This consideration did not play a role in this case.

C	D	E
First disinvested, later reimbursed again	**Reimbursement maintained**	**Reimbursement maintained**
Respondents suggested that, because health insurers would not have much financial benefit from reimbursement (i.e. the health benefits of the case intervention), they were not for reimbursement, although they were also not strongly against reimbursement.	Respondents described that health insurers did not have any financial risk in this case because of the policy rules that were in place. According to respondents, this may have contributed to the lack of action undertaken by health insurers. Respondents also described that manufacturers had strong financial interests in the case. For this reason, manufacturers have tried to exert pressure.	NA[1]
Respondents described that effectiveness only played a role in reversing disinvestment.	According to respondents, lack of evidence of effectiveness combined with limited effectiveness (and very high costs) were the main reasons to consider disinvestment. However, respondents described that lack of evidence of effectiveness was merely an indicator of the need for additional research, not a reason for disinvestment.	Respondents described that there was a lack of scientific evidence of effectiveness. However, there was broad consensus that case stories and qualitative data was sufficient to determine adequacy and soundness of the case intervention. For this reason, effectiveness does not appear to have played a large role.
This consideration did not play a role in this case.	According to respondents, lack of evidence of effectiveness combined with limited effectiveness and high costs, resulting in a very unfavourable cost-effectiveness ratio, were the main reasons to consider disinvestment. However, respondents described that there was a lack of societal support for a role of cost-effectiveness in reimbursement decisions. Therefore, the role of cost-effectiveness was limited. Despite this, respondents considered price negotiations essential for reimbursement to be maintained.	This consideration did not play a role in this case.

Themes	**Cases**	
	A	**B**
	Fully disinvested	**Partially disinvested**
- Necessity[2]	These considerations did not play a role in this case.	Respondents described that whether it would be feasible for patients to pay the case interventions themselves, was an important reason for disinvestment and played a major role in determining the exceptions (i.e. patient groups for whom reimbursement would be maintained). Furthermore, whether treatment was considered medically necessary was also very important in determining the exceptions.
- Feasibility[3]	These considerations did not play a role in this case.	Respondents described that the large budget-impact played a role in this case. Furthermore, they described that the administrative burden of making exceptions to disinvestment played a major role to determine the exceptions made (i.e. patient groups for whom reimbursement would be maintained).
- Other[4]		

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[1] NA: not applicable. Nothing with regards to this theme was addressed in the interviews.

[2] Necessity covers both the medical necessity of an intervention and the necessity to insure an intervention. Medical necessity covers aspects such as disease burden, rule of rescue and whether there is an alternative treatment available. Necessity to insure covers aspects such as 'who is responsible for paying for the intervention' and 'can patients afford to pay for the intervention themselves'. See for more information the paper of Kleinhout-Vliek et al: Kleinhout-Vliek T, de Bont A, Boer B. The bare necessities? A realist review of necessity argumentations used in health care coverage decisions. Health Policy. 2017;121(7):731-44.

[3] Feasibility covers aspects such as budget-impact (i.e. is paying feasible?), indication and administration (e.g. administrative burden, feasibility of regulations), and organization of care (e.g. can the intervention be (de-)implemented in the current organization of care?).

[4] This covers considerations that played a major role but are beyond the formal package criteria. If no other considerations played a role, this cell is left blank.

C	D	E
First disinvested, later reimbursed again	**Reimbursement maintained**	**Reimbursement maintained**
According to respondents, whether patients can pay for the case intervention themselves, and individual and societal responsibility to pay for the case intervention, were the main arguments in the discussion on reimbursement. Other necessity aspects that played a smaller role were the disease burden and the large number of patients.	Respondents described that medical necessity of the case interventions was evident, because of disease severity, lack of alternative treatment and the small number of patients. This contributed to maintaining reimbursement.	Respondents described that whether patients can afford to pay for the case intervention themselves and individual and societal responsibility to pay for the case intervention, played a large role in policy development and decision-making in this case.
Respondents described that budget-impact only played a role in reversing disinvestment. Other feasibility considerations did not play a role in this case.	These considerations did not play a role in this case.	According to respondents, feasibility considerations, such as budget impact, and organization and registration of care have also played a role.
		Respondents described that solidarity considerations were important in agenda-setting.

3

Societal Views In The Netherlands On Active Disinvestment Of Publicly Funded Healthcare Interventions

Rotteveel AH
Reckers-Droog VT
Lambooij MS
de Wit GA
van Exel NJA

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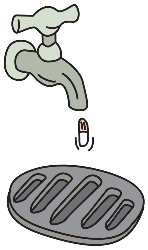

Abstract

Objectives: To obtain public support for the active disinvestment (i.e. policy decision to stop reimbursement) of healthcare interventions, it is important to have insight in what the public thinks about disinvestment and which considerations they find relevant in this context. Currently, evidence on relevant considerations in the disinvestment context is limited. Therefore, this study aimed to explore the societal views in the Netherlands on the active disinvestment of healthcare interventions and obtain insight into the considerations that are relevant for those holding the different views.

Methods: A Q-methodology study was conducted among a purposively selected sample of citizens (n=43). Data were collected in June and July 2019. Participants individually ranked a set of 43 statements broadly covering the issues that participants could consider relevant in the disinvestment context, from 'least agree' to 'most agree'. Qualitative feedback on the statement ranking was collected from each participant using a questionnaire. Principal component analysis followed by oblimin rotation was used to identify clusters of participants with similar statement rankings. These clusters/factors were interpreted as distinct viewpoints using the factor arrays and qualitative questionnaire responses of participants.

Results: Four viewpoints were identified. People holding viewpoint I believe that reimbursement of necessary healthcare should be maintained, irrespective of its costs. People holding viewpoint II agree with viewpoint I, although they believe that necessity should be objectively determined. People holding viewpoint III think that unnecessary, ineffective and inefficient healthcare should be disinvested. People holding viewpoint IV, consider it most important that disinvestment decision-making processes are transparent and consistent.

Conclusion: Insight in the distinct viewpoints identified in this study contributes to a better understanding of why it has been considered difficult to obtain public support for disinvestment of healthcare interventions, and can help policymakers to change their approach to disinvestment to increase public support.

Introduction

The continuing rise in healthcare expenditure in OECD countries [1], has put increasing pressure on public financing of healthcare [2, 3]. To curb the growth in healthcare expenditure, policymakers have increasingly been considering disinvestment of healthcare interventions as a policy option [4, 5]. Two types of disinvestment can be discerned: 1) passive disinvestment, which is not dependent on any direct intervention by policymakers, but mainly results from changing practices of healthcare providers or withdrawal from the market by the manufacturer, and 2) active disinvestment, which is the full withdrawal, retraction, restriction or substitution of resources from certain existing healthcare interventions, as a result of policy decisions, affecting the accessibility of these interventions to patients [5-8]. There may be several reasons for the disinvestment of healthcare interventions, such as harm, limited effectiveness or not enough value for money [7]. Disinvestment may be the outcome of Health Technology Reassessment (HTR), which is the structured, evidence-based assessment of healthcare interventions, currently being used in the healthcare system, based on their clinical, economic, social and ethical aspects [9, 10].

The disinvestment of healthcare interventions has been described in the international literature as very difficult [10-12]. Even in cases where there was strong evidence that the intervention was not (cost-)effective, withdrawal of this intervention was considered a delicate issue [10]. Unsurprisingly, a review of empirical evaluations of disinvestment initiatives from several different countries concluded that only a limited number of these initiatives was successful [13]. Support from healthcare professionals, politicians, and the general public has been described as essential for successful disinvestment [6, 14]. To obtain public support, it is important to have insight in how the public feels about disinvestment and which considerations they find relevant in this context. Furthermore, the engagement of public preferences in disinvestment decisions has been described to have a range of benefits: 1) instrumental benefits such as improving decision outcomes, 2) democratic benefits such as citizen engagement, and 3) educational benefits such as raising public awareness on the complexity of disinvestment decisions [5].

However, the scientific literature on the considerations that citizens find relevant in the context of disinvestment is limited to two studies looking at disinvestment in the context of a specific intervention (i.e. cancer drugs and vitamin B12/folate pathology testing) and one study aiming to answer a specific question in the context of disinvestment (i.e. whether people would like to know if their care is rationed). These studies found that citizens consider it important that disinvestment decisions are transparent [15] and based on clear and consistent principles and real-world (cost)effectiveness data [16]. Furthermore, the latter study also found that citizens consider it important not to disrupt current treatments of patients [17]. Finally, in the context of testing, Street et al. found it to be important to take costs, alternatives, disease severity, accuracy, potential to benefit and externalities into account in disinvestment decision-making [18]. Although these studies already provided some insight in the considerations that citizens find relevant in the context of disinvestment, it seems relevant to also investigate the relevant considerations for disinvestment in general, instead of in a specific context, to be able to improve disinvestment decision-making and increase public support for these decisions. Furthermore, it also seems relevant to assess whether views on which considerations are relevant differ between citizens. Therefore, in this study, we aimed to explore the societal views on the active disinvestment of healthcare interventions and to identify the considerations that people holding the different views consider important in this context. To this end, we have used Q-methodology, a method combining aspects of quantitative and qualitative research methods that enables the elicitation of rich, holistic viewpoints on a certain topic in a certain population [19]. In this study, we focus on the active disinvestment of healthcare interventions, with healthcare interventions indicating a broad range of curative care, including medicines, therapies, surgeries, medical devices, services and other types of curative care.

We have explored the societal views on active disinvestment and relevant considerations in the context of the Dutch social health insurance system. In the Netherlands, the coverage of the basic health insurance package is determined by the Minister of Health, Welfare and Sports based on advice from the Ministry of Health, Welfare and Sports and the National Healthcare Institute. In its advice, the National Healthcare Institute assesses healthcare interventions on four criteria: effectiveness, cost-effectiveness (i.e. health effects in relation to the costs of the intervention), necessity (i.e. do disease severity and the costs per patient justify coverage), and feasibility (i.e. is coverage feasible) [20]. In policy documents, it is assumed that when a healthcare intervention does not meet these four criteria anymore, it will no longer be delivered by healthcare providers (i.e. passively disinvested) [21]. However, in the past, also several decisions have been taken to (partially) stop reimbursement (i.e. active disinvestment) [14].

Methods

Approach

In this study, we used Q-methodology because this method fits the explorative nature of this study very well. Through the combination of qualitative and quantitative research methods, Q-methodology enabled us to obtain rich, holistic descriptions of the societal viewpoints on active disinvestment in a systematic way [19].

In a Q-methodology study, participants are asked to rank a comprehensive set of statements on a grid, according to how they feel about these statements (e.g. agree/disagree, important/unimportant), and explain the motivation behind their rankings. By-person factor analysis is used to identify clusters of participants with highly correlated rankings of the statements. The interpretation of the factors is facilitated by the weighted average ranking of the statements for the participants statistically significantly and uniquely associated with each identified factor, together with the qualitative data obtained from these participants, when explaining their ranking of the statements [22, 23]. Q-methodology has been widely applied in the context of health, healthcare, and healthcare priority setting [19, 24-27].

Statement set development

We developed a statement set that was broadly representative for our topic of interest, i.e. the active disinvestment of publicly funded healthcare interventions, in three consecutive steps. In the first step, we aimed to derive a broad, comprehensive collection of considerations potentially relevant in the context of active disinvestment. To this end, we adopted the conceptual framework from our previous study in which stakeholders (i.e. policymakers, patients, healthcare providers and other stakeholders) were interviewed to obtain insight into active disinvestment processes and aspects determining their outcome. This conceptual framework consisted of the actors, considerations and structures that may play a role in the different stages of a disinvestment process [14]. Subsequently, the considerations from this framework were complemented by the considerations that are relevant for citizens as identified in three previous Dutch Q-methodology studies in the context of investment [19, 28, 29], and in the three previously published studies in the context of disinvestment [15-18]. This resulted in a broad framework containing 87 potentially relevant considerations (see Appendix A).

Secondly, in an iterative process, the augmented framework was critically reviewed by three researchers (AR, ML and JE) to merge duplicate/comparable considerations and to discard considerations that were not relevant given the topic of interest. For each of the remaining considerations, a statement was formulated by AR and critically reviewed by ML and JE to ensure that the statements were concise and clear. This process resulted in a set of 45 statements.

Thirdly, to test the comprehensiveness and clarity of the statement set (and the other study materials), a pilot test was conducted among a convenience sample (n=6). As a result of the pilot test, the statement "If it is difficult to do research after the effect of the treatment, reimbursement may be stopped" was removed from the statements set, as participants did not understand why it may be difficult to do research, a problem we could not resolve with additional explanation or rephrasing of the statement. The statement "It is important that all those involved should be consulted in decisions on stopping the reimbursement" was also removed as participants did not find this realistic. Based on the feedback provided by participants, we clarified the wording of six statements (i.e. statements 5, 10, 11, 12, 13, 18 and 34), the information letter, and instruction for participants. The final set of 43 statements and its embedding in the conceptual framework is presented in Appendix B. More details on statement set development (in Dutch) are available from the authors upon request.

Data collection

For efficiency reasons, data were collected in group sessions with, on average, seven participants each. In these sessions, held in June and July 2019, participants were instructed to conduct the tasks individually. Participants were recruited through a commercial panel company. This panel company approached a large sample for participation in this study by sending them the participant information letter. Participants who were willing to participate in this study, could subscribe to one of the scheduled group sessions. The panel company controlled the enrolment of subscribed participants in the group sessions to ensure that the sample was diverse with regards to age (≥ 18 years), education level, political affiliation, and geographical spread. We used these variables as sampling variables because we expected these to be predictive of people having different views on disinvestment, enabling us to include a purposive sample. Participants of the group sessions received a show-up fee of €42.50 to compensate them for their time and travel expenses. The group sessions were led by the same researcher (AR), alternately helped by one of the other authors.

Participants were recruited until data saturation was reached, that is when no new viewpoints emerged from the data [30]. Saturation was determined based on participants' statement rankings and their written explanation of their ranking and their viewpoint on disinvestment. To ensure that we interpreted the written explanations right, these were checked with participants during the group sessions. In determining saturation, we focused on the most characterizing statements of the ranking (i.e. those ranked highest and lowest) in combination with the provided explanations. This combination allowed us to develop a general understanding of the viewpoints of participants, allowing us to determine saturation before formal analysis was started.

After obtaining written informed consent, participants received a short oral explanation of the task. Subsequently, participants were handed over a written instruction, the sorting grid (see Figure 1), 43 randomly numbered cards containing the statements, and a glossary explaining terms that participants may consider difficult (i.e. basic benefits package and medical guidelines). The written instruction described the background of the task (see Appendix C), and asked participants to, first, read all statements and sort them into three piles (i.e. agree, disagree, neutral/don't know), then, to rank the statements from each pile onto the sorting grid (starting with the agree pile, followed by the disagree pile and, lastly, the neutral/don't know pile), and, finally, to check and confirm the overall ranking of the statements. After finishing the ranking exercise, participants completed a short questionnaire (see Appendix D) that asked them 1) to explain why they agreed most/least with the four statements placed in the outer columns of the sorting grid, 2) to phrase their opinion on the topic of interest in their own words, and 3) two questions about their current health (i.e. the EQ-5D-5L and the EQ-5D visual analogue scale (VAS) [31, 32]) because we hypothesized that participants' health may affect their view on disinvestment.

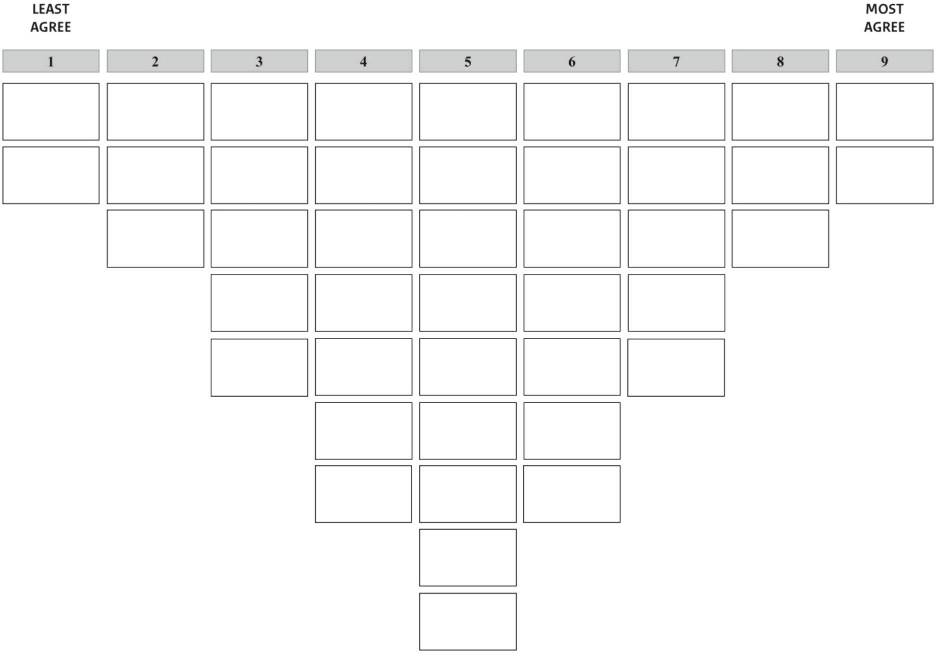

Figure 1: sorting grid used in this study

Ethics

Ethical approval was obtained from the internal review board of the Erasmus School of Health Policy & Management (IRB 2019-03). Written informed consent was obtained from all participants at the start of the task. Participants were informed that participation was voluntary and anonymous, and that they had the possibility to retract their consent at any time without having to give a reason. The research team did not have access to participants' contact information and handled the remaining participant information confidentially.

Data analysis and interpretation

A principal component analysis followed by oblimin rotation (a general form of oblique rotation [33]) was conducted to identify clusters of participants with highly correlated statement rankings. We determined the best number of factors from all possible factor solutions based on the following criteria: 1) Eigen Value of each factor >1.00; 2) a minimum of two non-confounded exemplars per factor (i.e. participants whose statement ranking was statistically significantly (i.e. $p<0.05$) and uniquely (i.e. square of factor loading > sum of square of the loading on other factors) associated with the factor); 3) a low or moderate correlation between the factors in the solution (with <0.30 being low, between 0.30 and 0.50 being moderate, and >0.50 being high [34]); and 4) coherence and distinctiveness of the interpretation of the factors as determined by the researchers (AR, VR and JE).

For all factors, a factor array was computed. Factor arrays concern an average ranking of the statements by participants who are statistically significantly and uniquely associated with the respective factor, weighted by their factor loadings, and represent how a hypothetical person with a correlation of 1.00 with that factor would have ranked the statements. Factor interpretation was based on the factor arrays, with special attention for the characterizing and distinguishing statements, and the qualitative questionnaire responses obtained from participants. Characterizing statements are those statements that are ranked the highest and lowest in the factor array, i.e. the statements participants associated with the viewpoint agreed least or most with. Distinguishing statements are those that have a statistically significantly different position in the factor array as compared to all other factors (p-value<0.05).

One participant placed 11 more statements on the ‘agree side (column 6-9)’ of the fixed sorting grid than was indicated. As the participant confirmed the statement ranking after having been pointed to the deviation from the intended form and the statement ranking was substantiated by the qualitative questionnaire responses, we decided to retain this participant in our analysis. To be able to include this ranking in our analysis, we analysed the data as a non-forced distribution. A sensitivity analysis excluding this participant and analysing the data as a forced distribution showed that the decision to retain this participant in our analysis had no significant effect on the outcome of the analysis.

The ‘qmethod’ package in Rstudio 1.2.1335 was used for the analyses [35, 36].

For the presentation of the study and our results in this manuscript, all statements, all presented quotes and the study material presented in the appendices were translated from Dutch to English by a professional translation company.

Results

Based on the statement rankings and the qualitative questionnaire responses, we found that saturation was reached after 43 participants. The mean (SD) age of the participants was 48.2 (16.4) years and the majority was male (56%, see Table 1). Participants were well distributed across education levels and well spread across the Dutch political spectrum. Geographical spread was ensured by the conduct of data collection on different locations across the Netherlands.

Table 1: Descriptive statistics of the participants (N=43)

		N (%)	Mean (SD[a])	Median (quartiles)
Sampling characteristics				
Age			48.2 (16.4)	54.0 (35.0-59.0)
Gender	Males	24 (56)		
	Females	19 (44)		
Education level[b]	Low	14 (33)		
	Middle	13 (30)		
	High	16 (37)		
Political spectrum[c]	Left	18 (42)		
	Centre	8 (19)		
	Right	14 (33)		
	Missing	1 (2)		
Other characteristics				
Quality of life	EQ-5D-5L utility value (0-1)[d]		0.84 (0.15)	0.89 (0.82-0.92)
	EQ-5D-VAS (0-100)		79 (17)	80 (70-90)
Living situation	Alone	10 (23)		
	Alone, but with children	4 (9)		
	Together with partner	14 (33)		
	Together with partner and children	10 (23)		
	With parents	5 (12)		
Children	No	15 (35)		
	Yes	28 (65)		

[a] SD= standard deviation

[b] Education levels correspond to the SOI 2016 and the ISCED 2011 classifications

[c] The parties participants would vote if there would be elections now were categorized by the commercial panel in left, centre and right. We adopted this categorization to report on this variable here, as the specific party participants would vote is not informative for international readers. However, when checking for spread across the political system, we looked at the specific parties the participants would vote, not the categorization as reported here.

[d] Calculated from the EQ-5D-5L score using the Dutch tariff [37]

Based on the statistical criteria described in the method section, a maximum of four factors was supported by the data. After a first inspection, all four factors were retained as distinct viewpoints, as each of them had a coherent and clear interpretation and seemed to represent a distinct viewpoint on disinvestment. The factors were defined by 19, 4, 12 and 3 participants, respectively, and Eigen Values were between 2.8 and 9.1. Five participants were not statistically significantly associated with one of the factors, because they loaded on multiple factors. Together, the factors explained 48% of the data variance, with 6.5% to 21.1% of explained variance per factor. Correlations between the four factors were low for factors 1 and 2 (0.27), for factors 1 and 3 (0.22), for factors 2 and 3 (0.21), and for factors 2 and 4 (0.21). Correlations were moderate for factors 1 and 4 (0.34), and for factors 3 and 4 (0.45). The factor loadings of participants are displayed in Appendix E.

Table 2 shows the factor arrays. The factor arrays display the weighted average ranking of the statements by all participants who are statistically significantly and uniquely associated with the factor: a score of +4 indicates that these participants would rank the statement in the most agree column of the sorting grid in Figure 1 (column 9), a score of -4 indicates that these participants would rank the statement in the least agree column of the sorting grid (column 1), a score of 0 indicates that these participants would rank the statement in the middle of the sorting grid (column 5). An asterisk displayed alongside the score indicates that the ranking of this statement is distinguishing between that factor and all other factors.

Table 2: Factor arrays

#	Statement	F1[a]	F2[a]	F3[a]	F4[a]
1	If the treatment is effective, reimbursement should not be discontinued.	+2*	+1	0	+4*
2	If the treatment leads to small health benefits, reimbursement may be discontinued.	-3*	0	0	-1*
3	If the quality of life of patients is still poor after treatment, reimbursement of this treatment may be discontinued.	-4*	+2*	+0*	-2*
4	If the quality of life of patients is good without the treatment, reimbursement of this treatment may be discontinued.	-1	-1	+2	+1
5	If there is a significant difference in the effect of the treatment between patients, reimbursement of the treatment should only be discontinued for patients in whom it has little effect.	-1	-3*	0	0
6	While it is not yet clear which patients will benefit from the treatment, this treatment should continue to be reimbursed for all patients.	0	0	-1	0
7	If the chances of the treatment having an effect are small, the reimbursement may be discontinued.	-3	-1	0	0
8	If the effect of the treatment cannot be scientifically demonstrated, reimbursement for this treatment may be discontinued.	-1	-1	+2*	0

Table 2 continued:

#	Statement	F1[a]	F2[a]	F3[a]	F4[a]
9	While research into the effect of the treatment is still ongoing, the reimbursement should not be discontinued	+1*	-4*	-1*	-2*
10	If the treatment is included in the medical guideline, reimbursement for this treatment should not be discontinued.	+2	+1	0	0
11	If doctors believe that patients are benefiting from the treatment, reimbursement should not be discontinued.	+3*	0	+2	+1
12	If patients feel they are benefiting from the treatment, reimbursement should not be discontinued.	0*	-1	-3*	-2
13	If the treatment costs are high in relation to its effects, reimbursement may be discontinued.	-2*	0	+1*	0
14	If a cheaper alternative to the treatment is available (which works equally well), reimbursement of the treatment may be discontinued.	0	0	+3	+2
15	If the treatment is medically necessary, reimbursement may not be discontinued.	+4	+4*	+2	+3
16	If a serious illness is concerned, reimbursement for its treatment should not be discontinued.	+4*	+1	+1	-1
17	If a chronic illness is concerned, reimbursement for its treatment should not be discontinued.	+2	+1	+1	+2
18	If patients feel that the treatment is necessary, its reimbursement should not be discontinued.	0*	-3	-4*	-3
19	If a particular complaint is part of normal life, reimbursement for its treatment may be discontinued.	-1	0	+4*	-4*
20	If patients can pay for the treatment themselves, reimbursement for this treatment may be discontinued.	-2*	+2*	-1*	-4*
21	If the patient is personally responsible for developing a condition, reimbursement for its treatment may be discontinued.	-2	-2	0*	-2
22	If the treatment is frequently used just because it is reimbursed, reimbursement may be discontinued.	-1*	0	+4*	+2
23	If the treatment helps patients to maintain their dignity, reimbursement should not be discontinued.	0	-2	-1	0
24	If the treatment is the only treatment option for a condition, its reimbursement should not be discontinued.	+3	+4	+1	+2
25	If a small patient group is concerned, reimbursement for the treatment of this patient group should not be discontinued.	+1	-2	-2	0
26	If a condition is stressful for the patients' family, reimbursement for its treatment should not be discontinued.	-1	-1	-2	-3*
27	If a contagious condition is concerned, reimbursement for its treatment should not be discontinued.	+1	+2	+3	0
28	If the treatment improves patients' participation in society, its reimbursement should not be discontinued.	+1*	+3	0*	+3
29	If the reimbursement of a treatment is discontinued, patients who are already being treated with this treatment should still have it reimbursed.	0[b]	+1[b]	0[b]	+1[b]
30	If there is no support in society for discontinuing reimbursement of the treatment, the reimbursement should not be discontinued.	0	+3*	-1	-2*
31	If a disproportionately large part of the care budget is spent on the treatment, its reimbursement may be discontinued.	-3*	-1	-2	+1*

Table 2 continued:

#	Statement	F1[a]	F2[a]	F3[a]	F4[a]
32	If the care is organized in such a way that the treatment cannot be offered safely, its reimbursement may be discontinued.	-1	-2	+1	+1
33	When deciding to discontinue reimbursement, the same criteria should always be applied in the same manner.	-2	-2	+1*	+4*
34	When deciding to discontinue reimbursement, it should always be made clear how and based on which criteria the decision was made.	0*	+2	+1	+3*
35	If the discontinuation of the reimbursement of the treatment disproportionately affects a certain group of people in society, this reimbursement should not be discontinued.	+1	+1	-2*	+1
36	If the discontinuation of the reimbursement of the treatment concerns a vulnerable group in society, this reimbursement should not be discontinued.	+1	+2	-3	-1
37	When it comes to treatment of a life-threatening condition in young people, reimbursement should not be discontinued.	+3	-1*	+3	+1*
38	Because it is impossible to express a human life in terms of money, costs should not play a role in decisions about discontinuing reimbursement.	+1*	+3*	-3	-3
39	When it comes to care for patients who have a short time left to live, reimbursement of this care should not be discontinued	0	0	-1	-1
40	It is morally wrong to deny patients existing treatment.	+2*	-4*	-2	-1
41	If the treatment can prevent people from becoming ill, its reimbursement should not be discontinued.	+2	+1	+2	+2
42	If the costs of the treatment per patient are high, reimbursement may be discontinued	-4*	0	-1	-1
43	If the healthcare provider has not yet recouped its investment in the treatment, its reimbursement should not be discontinued.	-2	-3	-4*	-1*

[a] The numbers displayed in these columns represent the factor score of each statement in each factor. This score indicates the column of the sorting grid (figure 1) where the statement would be placed if it would be sorted by a person with the particular view corresponding with the factor, with -4 indicating the most left column (least agree), 0 indicating the middle column and +4 indicating the most right column (most agree).
[b] Consensus statement (no difference between factors at a p-value <0.05)
*Distinguishing statement (p-value <0.05)

Below, the interpretation of the four factors as four distinct viewpoints on what people consider important in disinvestment decisions is presented. The viewpoints are described using the characterizing and distinguishing statements for that factor (where # represents the statement number, with the corresponding factor array score, and * indicates that the statement is distinguishing). Quotes selected from the qualitative questionnaire responses of participants statistically significantly and uniquely associated with the factor (with participant ID between brackets) are used for illustration. These quotes were selected based on how well they illustrated our findings.

Viewpoint I

People holding this viewpoint consider it important that the reimbursement of necessary healthcare will be maintained. They believe healthcare is necessary if it concerns treatment for severely ill patients (#16, +4*; #37, +3), if a treatment is included in medical guidelines (#10, +2), if physicians think patients will benefit from the treatment (#11, +3*) and if the treatment is considered medically necessary (#15, +4): *"If there are medical reasons that have been determined by a doctor, this should always be reimbursed!" (ID-33).* Such treatments should remain in the basic benefits package, even if these only result in small health gains (#2, -3*; #7, -3) or in a low quality of life after treatment (#3, -4*). People holding this viewpoint consider it morally wrong to deny patients an existing treatment (#40, +2*), in particular if no alternative treatment is available to patients (#24, +3), as is illustrated by the following quote: *"As the text [statement] indicates. From a moral point of view, I do not think it is acceptable to deny existing treatment for any reason whatsoever (probably financial reasons)." (ID-01)* People holding this viewpoint believe that the costs (#42,-4*), cost-effectiveness (#31, -3*) and budget-impact (#31, -3*) of treatments should not play any role in disinvestment decisions, as is illustrated by the following quotes: *"That people get the care they need, no matter what it costs." (ID-01); "Denying the right/necessary care should not be allowed. The country is prosperous enough to be able to offer this to every citizen." (ID-20)* For further reference, we will call this viewpoint "Maintain necessary healthcare, even if it is expensive or only results in small health gains".

Viewpoint II

Similar to people holding viewpoint I, people holding this viewpoint believe that the reimbursement of necessary care (i.e. treatments that are medically necessary or are the only treatment option available) should be maintained (#24, +4; #15, +4*), irrespective of its costs (#38, +3*): *"Care should be accessible to everyone, especially if it is medically necessary." (ID-07)* However, they believe necessity should be objectively determined and not, for example, based on insufficient scientific evidence (#9, -4*) or patient opinion (#18, -3): *"Most important is if the treatment does not work after thorough study." (ID-21)* Furthermore, they believe that if quality of life remains low after treatment (#3, +2*), if providers have not earned back their investment (#43, -3), and if people can pay for their own treatment (#20, +2*) reimbursement may be stopped: *"The costs of care are so high that the premium system should be changed. Wealthy people can then pay more and the socially disadvantaged can pay less." (ID-21)* Hence, they believe that in some situations, denying patients treatment is not morally wrong (40, -4*). Furthermore, what is distinguishing for this viewpoint, is the importance attached to public support for disinvestment decisions. If there is no public support for stopping the reimbursement of healthcare, reimbursement should be maintained (#30, +3*): *"Everyone is entitled to care if there is no support [for stopping reimbursement]." (I08)* Also distinguishing for this viewpoint is the importance attached to participation in society and protecting vulnerable groups in society: when treatment contributes to patients being able to participate in society (#28, +3), or if stopping treatment would affect a vulnerable group (#36, +2), reimbursement should be maintained. For further reference, we call this viewpoint "Maintain necessary healthcare, if objectively determined and if there is no support for disinvestment".

Viewpoint III

People holding this viewpoint consider it important to spend the healthcare budget in a well-considered way. Therefore, they think that the reimbursement of healthcare that is not necessary or that does not have any effect can be stopped (#19, +4*; #22, +4*; #4, +2): *"Everyone has aches and pains sometimes. The idea is not that you need immediate care for every little ache and pain (that is part of normal life). This is at the expense of the people who really need care." (ID-04); "There can be all kinds of reasons to deny patients existing treatments. Maybe a specific treatment does not work in their case, or the improvement in health would not outweigh the suffering that this treatment entails. So it is not morally wrong, but there has to be a logical reason for denying a patient certain treatment." (ID-36)* Furthermore, the reimbursement of healthcare that is not effective (#8, +2*) or not cost-effective can be stopped as well (#14, +3; #38, -3*; #13, +1*): *"From a business point of view, it is right to make a financial consideration. If it can be done more cheaply with the same desired effect, then I think that's right." (ID-26)* Whether disinvestment affects a vulnerable group or any other group in society disproportionally should not be taken into account in disinvestment decisions (#35, -2*; #36, -3). If a treatment would prevent other people from becoming ill (#41, +2), is targeted at contagious diseases (#27, +3) or concerns a life-threatening disease in younger people (#37, +3), than that would be a good use of the healthcare budget and, hence, reimbursement can be maintained. People holding this viewpoint think that the patients' voice should not play a role in disinvestment decisions. Whether patients feel they benefit from treatment or consider the treatment necessary is not relevant (#18, -4*; #12, -3*): *"I think the doctor or hospital should decide that, and not the patients themselves." (ID-15)* Furthermore, people holding this viewpoint disagreed most with the statement that treatment should remain reimbursed until healthcare providers earned back their investment (#43, -4*): *"Nonsense, the composition of the basic health insurance package should never serve to balance the cash book of the pharmaceutical industry. This industry is already doing enough itself, with that argument to raise prices absurdly." (ID-41)* For further reference, we call this viewpoint "Disinvest unnecessary, ineffective and inefficient healthcare".

Viewpoint IV
People holding this viewpoint consider it important that disinvestment decision-making (i.e. both the process and the considerations underlying the decision) is transparent and consistent. If reimbursement for a certain treatment is stopped, it should be clearly explained why (#34, +3*). Furthermore, disinvestment decisions should always be based on the same set of criteria (#33, +4*). Why transparency and consistency are considered important is explained in the following quotes: *"Because everyone should receive the same treatment and opportunities." (ID-29); "Then it's easier to understand why the decision to discontinue it [reimbursement] has been taken." (ID-29)*

In this view, effectiveness (#1, +4*) and medical necessity (#15, +3) are important criteria for care to remain reimbursed, particularly if it helps people to better function in society (#28, +3). However, the sustainability of the healthcare system is also a matter of concern: *"The sustainability of the care system is very important and this means that the right balance must be found between good healthcare and financial sustainability. One should not lose sight of the financial picture." (ID-12)* Therefore, the costs and budget-impact of a treatment should be considered as well (#31, +1*, #38, -3): *"Discontinuation may be possible when costs are unnecessarily high and the same result is achieved with other cheaper means." (ID-38)*

If disinvestment decisions are made in a transparent and consistent way, (a lack of) public support (#30, -2*), patient preferences (#18, -3*), the burden of the disease on the patient (#16, -1) and the patient's family (#26, -3*), or whether people can pay for the treatment themselves (#20, -4*) should not lead to exceptions to the rules. Also, whether a complaint is part of normal life should not play a role in disinvestment decision-making (#19, -4), because this cannot be determined in a clear-cut, consistent manner. For further reference, this viewpoint is described as "Transparent and consistent disinvestment decision-making processes".

Discussion

The aim of this study was to explore the societal viewpoints in the Netherlands on the active disinvestment of healthcare interventions and to identify the considerations that people holding these viewpoints find relevant in this context. Four distinct viewpoints were identified, which can be shortly described as: 1) Maintain reimbursement of necessary healthcare, even if it is expensive or only results in small health gains; 2) Maintain reimbursement of necessary healthcare, if objectively determined or if there is no public support for disinvestment; 3) Disinvest unnecessary, ineffective or inefficient healthcare; and 4) It is most important that disinvestment decision-making processes are transparent and consistent.

Comparison of our findings with the disinvestment literature

There are several similarities between the findings of the three previous studies on relevant considerations in the context of disinvestment and some of the viewpoints identified in our current study. For instance, in our current study, we found that people holding viewpoint 4 consider it important that disinvestment decision-making processes are transparent and consistent. This seems to confirm the finding of Bentley et al. that citizens consider it important that disinvestment decisions are based on clear and consistent principles [16] and the finding of Coast et al. that people consider it important that disinvestment decisions are transparent [15]. Furthermore, some of the relevant considerations identified by Street et al. [18] are supported by some of the viewpoints identified in our study. For instance, taking costs and effectiveness into account in disinvestment decisions is supported by viewpoints 3 and 4. Moreover, taking the availability of alternative treatments into account is supported by viewpoints 1 and 2. However, it becomes clear from our current study that these considerations are not supported by all viewpoints, indicating that it is important to take heterogeneity in citizens' viewpoints into account.

Secondly, there are also some differences between the findings of previous studies and the findings of the current study. For instance, although Costa et al. found that citizens are reluctant to disrupt treatment of current patients [17], this is not supported by any of the viewpoints identified in our current study: Statement 29 on continuing reimbursement for current patients was ranked in the middle of the distribution for all viewpoints. Even though this does not necessarily mean that all participants sorted this statement in the middle of the sorting grid (i.e. agrees and disagrees among people holding the viewpoints may also cancel each other out for this statement), it does mean that in our current study we did not identify any viewpoint that found this consideration relatively important compared to the other considerations.

Comparison of our findings with the decision criteria currently used in Dutch policy

In the Netherlands, the National Healthcare Institute uses the four criteria effectiveness, cost-effectiveness, necessity and feasibility to advise the Ministry on the reimbursement of healthcare interventions. With regards to investment decisions, effectiveness is a 'knock-out' criterion [20]. However, our study shows that support for effectiveness as a criterion for disinvestment decisions varies. People holding viewpoint 1 consider effectiveness relatively unimportant, while people holding viewpoint 2 rank these considerations in the middle of the sorting grid, and people holding viewpoints 3 or 4 consider effectiveness relatively important. A similar pattern can be observed for the cost-effectiveness criterion: people holding viewpoint 1 or 2 think cost-effectiveness should not play a role, while people holding viewpoint 3 or 4 consider this criterion important. However, there seems to be some consensus on the importance of the medical necessity criterion: statement 15 on medical necessity is located in the most agree tail of the distribution for all viewpoints (+4, +3, +2, +4). Nonetheless, there seem to be some differences between the viewpoints on the consequences this criterion should have: people with viewpoint 1 or 2 generally do not see a lack of medical necessity as a reason to disinvest an intervention or service, while people holding viewpoint 3 or 4 indicate that non-necessary care should be disinvested. Furthermore, from the factor arrays some differences with regards to the interpretation of medical necessity can be observed. For instance, people holding viewpoint 2, 3 or 4 generally do not agree with taking the patient's opinion on necessity into account (statement 18), while people holding viewpoint 1 are more open to this. Moreover, while people holding viewpoint 3 think that treatments for illnesses that are part of normal life should be disinvested (statement 19), people holding viewpoint 4 do not agree with this (as this cannot be determined in a clear-cut way) and people holding viewpoint 1 or 2 are more neutral with regards to this statement. Finally, there is no consensus on the relative importance of the feasibility considerations budget-impact and public support, included in this study. Viewpoint 3 and 4 support the use of budget-impact as a criterion in disinvestment decisions, while viewpoint 1 does not support this and viewpoint 2 is neutral. Furthermore, viewpoint 2 considers public support very important, while viewpoint 1 does not seem to have a strong opinion on this and viewpoint 3 and 4 do not consider it to be important.

Methodological considerations

The use of Q-methodology in this study allowed us to obtain rich, holistic descriptions of the existing societal viewpoints on active disinvestment in the Netherlands [19]. Because of the combination of qualitative and quantitative research methods used in Q-methodology, it provided us with the opportunity to obtain in-depth insights in the existing viewpoints which are discerned in a systematic way.

However, Q-methodology also has three disadvantages compared to other research methods. Firstly, because of the structured way of data collection, by asking all participants to sort the same set of statements with the same instruction, it could be argued that Q-methodology is not able to obtain such rich insights as non-structured or semi-structured qualitative research methods can. However, the statement set used in this study was based on the findings of our previous study in which we obtained in-depth insight in the considerations that may be relevant in disinvestment decisions through a large number of semi-structured interviews [14]. Subsequently, the statement set was carefully developed and pilot-tested to ensure that the statements would cover the variety of aspects relevant in active disinvestment. After finishing statement sorting, participants had the opportunity to phrase their opinion. This process enabled us to evaluate whether any relevant aspect was missing. In this process no missing aspects were identified, indicating that all considerations relevant in the context of disinvestment were included in this study.

Secondly, because of the purposive sampling method, Q-methodology is not suited to examine how the different viewpoints are distributed across society and whether the viewpoints are statistically significantly associated with any socio-demographic characteristics [23, 38]. Survey approaches allowing a large number of probabilistically sampled participants to rate their agreement with aspects of the viewpoints (e.g. a selection of statements that discriminate best between viewpoints or short viewpoint descriptions) are more appropriate for this purpose [23, 38].

Finally, Q-methodology is also not the appropriate method to measure the relative strength of the preferences for the different considerations in disinvestment decision-making. For this purpose, choice experiments would be more suitable. The characterizing and distinguishing statements identified in this study can serve as input for attribute development in future choice experiments.

Strengths and limitations

To our knowledge, this is the first Q-methodology study examining the societal views on the active disinvestment of healthcare interventions. We ensured reliability of statement set development and viewpoint interpretation by conducting the development of the set of statements as well as the interpretation of the viewpoints with three researchers (i.e. triangulation) [39]. Furthermore, pilot testing of the study materials and the face-to-face approach to data collection ensured reliability of data collection [22].

Despite these strengths, the current study also has some additional limitations to the ones described in the previous section. Firstly, as for efficiency reasons the data were collected in groups, participants' explanations of statement sortings were collected using a questionnaire. This resulted in short, written explanations of participants' viewpoints. To ensure that the researchers would interpret these short explanations correctly, questionnaire responses were checked during the group sessions and any written clarification of the responses was requested if necessary. Despite this precaution, some nuances in viewpoint interpretation may have been overlooked due to the short, written viewpoint explanations provided by participants.

Secondly, this study has been conducted in a Dutch setting. As disinvestment processes are considered context-specific [10], researchers and policymakers are recommended to take the context into account when considering the implications of the results of this study in their context. However, as a previous Q-methodology study assessing allocation preferences in the investment context in nine European countries (i.e. Denmark, France, Hungary, the Netherlands, Norway, Poland, Spain, Sweden and the UK) only found small differences in the views between these countries [19], we believe that the views identified in this study are also broadly applicable to other (European) contexts.

Policy implications

Active disinvestment has been described as a delicate issue [10], with public support being considered essential for its success [6]. Even though it is also part of a policymaker's job to make unpopular decisions, such as disinvestment decisions, public support may increase the success of such decisions. The results of this study show considerable heterogeneity between groups in society in their views on disinvestment and in the considerations they consider relevant in this context. This indicates that it will be very difficult, if not impossible, to design the disinvestment process and corresponding communication in such a way that the preferences of all people holding the different viewpoints will be met. Hence, it indicates that it is very difficult to obtain support for disinvestment from all groups in society.

However, despite the identified heterogeneity between the viewpoints, the results of this study also provide policymakers with guidance on how to increase public support for disinvestment. Firstly, our study shows that all four viewpoints support the use of medical necessity as a consideration in disinvestment decisions. Therefore, in selecting candidate interventions for disinvestment and in the communication on disinvestment decisions, we recommend policymakers to focus on medical necessity. However, the interpretation and consequences attached to this criterion differ between viewpoints, possibly explaining the broad support for this statement. Despite of this, people from all viewpoints seem to support necessity as has been determined by healthcare professionals as a way to determine what medical necessity is. Therefore, policymakers are recommended to focus on this interpretation of medical necessity. Although people holding viewpoint 1 and 2 support medical necessity as a criterion, they only seem to see medical necessity as a reason to reimburse treatments, not as a reason to disinvest treatments. Therefore, to also increase support among people holding viewpoint 1 or 2, policymakers are recommended to put emphasis on the fact that disinvestment of non-necessary healthcare will create scope for the reimbursement of necessary healthcare.

Secondly, our study shows that people holding viewpoint 4 consider a transparent disinvestment process very important. Although transparency is less important for those holding the other viewpoints, these people still place this statement on the most agree side of the sorting grid, indicating that, to some degree, they agree with this statement. Therefore, we recommend policymakers to improve the transparency of the disinvestment process and corresponding communication to increase support from people holding viewpoint 4 and from people holding the other viewpoints as well.

Conclusion

In conclusion, this study shows that there are four distinct societal viewpoints on active disinvestment of publicly funded health technologies that highlight important considerations for decision-making in this context. Insight in these viewpoints can thus help policymakers to better understand why it has been considered difficult to obtain public support for disinvestment, and how this can perhaps be improved in the future.

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Appendices

Appendix A: First version conceptual framework

Domain	Consideration	Sub-consideration
Effectiveness	Common practice	
	Efficacy	
	Effectiveness	
	Size of the effect	
	Length of life vs quality of life	
	Variability	Heterogeneity
		Certainty of the effect occurring
	Strength of evidence	Amount of research
		Level of evidence
		Methodological quality
		Statistical significance
		Limitations in doing research
		Necessity for further research
	Clinical relevance	
	Patient relevance	
	Safety/adverse effects	
	Capacity to benefit	
	Mechanism of action	
Cost-effectiveness	ICER vs threshold value	
	Costs (per unit of the intervention)	
	Quality of life/QALYs	
	Uncertainty	
	Strength of evidence	Amount of research
		Level of evidence
		Methodological quality
		Limitations in doing research
		Necessity for further research
Necessity	Necessity to insure	Definition of illness
		Definition of treatment
		Individual Cost
		Individual responsibility/culpability
		Moral Hazard
		Range of Normality

Domain	Consideration	Sub-consideration
		Societal responsibility
		Amount of treatment received so far
	Medical Necessity	Medical Necessity
		Morbidity/Severity
		Need
		Rule of Rescue
	Other necessity aspects	Dignity
		Equity/fairness/justice
		Human Right
		(No) Alternative
		Patient-diagnosis
		Similar Treatments
		Number of Patients
		Societal impact
		Societal functioning
		Vulnerability/compassion
		Has been used to treat patients/ reimbursed before
		Age(ism) / fair innings
		Treatment unsuccessful so far
Feasibility	Support	
	Organization of care	
	Indication and administration	
	Financing	
	Legal and ethical	
	Consultation and anticipation	
	Commencing date	
	Consequences for healthcare consumption	
	Budget impact	Societal
		Health care budget
Other	Considered/fair process	Agenda-setting
		Policy-development
		Decision-making
		Implementation
	Healthcare delivery	Quality of care
		Customized healthcare
	Undesirable use	

Domain	Consideration	Sub-consideration
	Autonomy of healthcare providers	
	Freedom of choice/ autonomy for patients	
	Reliable government	
	Disappearance / marginalization disciplines	
	Signal of reimbursement	
	Values	Accessibility
		Solidarity
		Life is priceless
		Wrong to deny treatment
	Prevention (value of)	
	Emotional arguments	
	Financial arguments	Need to cut healthcare budget/ Finiteness of healthcare budget
		Profit/revenue
		Sunk costs
		Price setting/return on investment
	Distribution of healthcare benefits	
	Type of disinvestment	
	Budget allocation	

Appendix B: Final set of statements including its embedding in the conceptual framework

Domain	Considerations		Statements[1]	#
Effectiveness	Effect of treatment (general)	Effectiveness (general)	If the treatment is effective, reimbursement should not be discontinued.	1
		Size of the effect	If the treatment leads to small health benefits, reimbursement may be discontinued.	2
	Start- and end-point (quality of life before and after treatment)	Quality of life after treatment	If the quality of life of patients is still poor after treatment, reimbursement of this treatment may be discontinued.	3
		Quality of life before treatment	If the quality of life of patients is good without the treatment, reimbursement of this treatment may be discontinued.	4
	Variability	Heterogeneity	If there is a significant difference in the effect of the treatment between patients, reimbursement of the treatment should only be discontinued for patients in whom it has little effect.	5
		Uncertainty	While it is not yet clear which patients will benefit from the treatment, this treatment should continue to be reimbursed for all patients.	6
			If the chances of the treatment having an effect are small, the reimbursement may be discontinued.	7
	Scientific evidence	Evidence of effectiveness	If the effect of the treatment cannot be scientifically demonstrated, reimbursement for this treatment may be discontinued.	8
		Necessity for further research	While research into the effect of the treatment is still ongoing, the reimbursement should not be discontinued	9

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[1] The statements were translated from Dutch to English by a professional translation company

Domain	Considerations		Statements[1]	#
	Clinical practice	Guidelines	If the treatment is included in the medical guideline, reimbursement for this treatment should not be discontinued.	10
		Clinical relevance	If doctors believe that patients are benefiting from the treatment, reimbursement should not be discontinued.	11
	Patient relevance		If patients feel they are benefiting from the treatment, reimbursement should not be discontinued.	12
Cost-effectiveness	Value for money		If the treatment costs are high in relation to its effects, reimbursement may be discontinued.	13
	Incremental cost-effectiveness		If a cheaper alternative to the treatment is available (which works equally well), reimbursement of the treatment may be discontinued.	14
Necessity	Medical Necessity	Medical Necessity	If the treatment is medically necessary, reimbursement may not be discontinued.	15
		Burden of illness	If a serious illness is concerned, reimbursement for its treatment should not be discontinued.	16
			If a chronic illness is concerned, reimbursement for its treatment should not be discontinued.	17
		Need	If patients feel that the treatment is necessary, its reimbursement should not be discontinued.	18
	Necessity to insure	Normality of health complaint	If a particular complaint is part of normal life, reimbursement for its treatment may be discontinued.	19
		Individual Cost	If patients can pay for the treatment themselves, reimbursement for this treatment may be discontinued.	20

Domain	Considerations			Statements[1]	#
		Individual responsibility/ culpability/ lifestyle		If the patient is personally responsible for developing a condition, reimbursement for its treatment may be discontinued.	21
		Moral Hazard		If the treatment is frequently used just because it is reimbursed, reimbursement may be discontinued.	22
	Other necessity aspects	Dignity		If the treatment helps patients to maintain their dignity, reimbursement should not be discontinued.	23
		No alternative		If the treatment is the only treatment option for a condition, its reimbursement should not be discontinued.	24
		Number of Patients	Rarity	If a small patient group is concerned, reimbursement for the treatment of this patient group should not be discontinued.	25
		Societal impact	Burden/ spill over on patients' family	If a condition is stressful for the patients' family, reimbursement for its treatment should not be discontinued.	26
			Effect on others becoming ill	If a contagious condition is concerned, reimbursement for its treatment should not be discontinued.	27
			Productivity (social and societal participation)	If the treatment improves patients' participation in society, its reimbursement should not be discontinued.	28
		Has been used to treat patients/ been reimbursed before		If the reimbursement of a treatment is discontinued, patients who are already being treated with this treatment should still have it reimbursed.	29
Feasibility	Support			If there is no support in society for discontinuing reimbursement of the treatment, the reimbursement should not be discontinued.	30
	Budget impact	Health care budget impact		If a disproportionately large part of the care budget is spent on the treatment, its reimbursement may be discontinued.	31

Domain	Considerations		Statements[1]	#
	Safety		If the care is organized in such a way that the treatment cannot be offered safely, its reimbursement may be discontinued.	32
Other	Considered/ fair process	Consistency of criteria	When deciding to discontinue reimbursement, the same criteria should always be applied in the same manner.	33
		Transparency	When deciding to discontinue reimbursement, it should always be made clear how and based on which criteria the decision was made.	34
	Values	Equity/fairness/ justice/ solidarity	If the discontinuation of the reimbursement of the treatment disproportionately affects a certain group of people in society, this reimbursement should not be discontinued.	35
			If the discontinuation of the reimbursement of the treatment concerns a vulnerable group in society, this reimbursement should not be discontinued.	36
			When it comes to treatment of a life-threatening condition in young people, reimbursement should not be discontinued.	37
		Life is priceless	Because it is impossible to express a human life in terms of money, costs should not play a role in decisions about discontinuing reimbursement.	38
		End-of-life care	When it comes to care for patients who have a short time left to live, reimbursement of this care should not be discontinued	39
		Wrong to deny treatment	It is morally wrong to deny patients existing treatment.	40
	Prevention (value of)		If the treatment can prevent people from becoming ill, its reimbursement should not be discontinued.	41

Domain	Considerations		Statements[1]	#
	Costs/ price (per unit of the intervention)	Price/costs	If the costs of the treatment per patient are high, reimbursement may be discontinued	42
		Price setting/return on investment	If the healthcare provider has not yet recouped its investment in the treatment, its reimbursement should not be discontinued.	43

Appendix C: Background of the task for participants

In the Netherlands, the costs of healthcare will continue to increase. In order to keep healthcare accessible and affordable for everyone, policymakers are, therefore, critically examining whether new medical treatments should be included in the basic health insurance package. In the past, this was examined less critically. As a result, it is possible that the basic health insurance package currently includes treatments that we no longer want to reimburse. It is, therefore, important to also consider whether treatments can be removed from the basic health insurance package; in other words, whether the reimbursement of some treatments can be discontinued. Discontinuing reimbursement can create scope for reimbursing other, new treatments without increasing the cost of care. In this study, we examine what considerations citizens believe the government should take into account when making decisions about discontinuing certain healthcare reimbursements.

We will give you 43 cards with statements to read. These statements are about the role that various considerations may play in decisions about discontinuing reimbursement for treatments. We will ask you to sort these statements according to the degree to which you agree with them. We will then ask you to elaborate on your choices. You will also be asked a few closing questions. The study will take around an hour of your time. This study is all about your opinion; there are no right or wrong answers.

Appendix D: Questionnaire

Instruction: We kindly ask you to answer several questions. We are interested in your opinion, so there are no right or wrong answers. This questionnaire contains 7 questions, divided over 6 pages. Please read the questions carefully and try to answer every question.

1. Please take another look at the two statements you placed on the far right of the sorting grid, under "9". Please write down the numbers and explain why you MOST AGREE with these statements?

Card number: ……………….
Explanation:

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……
……
……

Card number: ……………….
Explanation:

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……
……
……
……

2. Please take another look at the two statements you placed on the far left of the sorting grid, under "1". Please write down the numbers and explain why you LEAST AGREE with these statements?

Card number: ……………….
Explanation:

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Card number: ………………

Explanation:

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3. Were there any statements that you did not understand? If so, please note the numbers of these statements below.

Card numbers: …………………………………………………………………………

4. Can you describe what is most important to you in deciding whether or not to discontinue reimbursement for a treatment?

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……

→ Question 5 is on the next page

→ Questions 5 and 6 are about your health. We are asking you about your health to find out how a person’s health influences how they sort the statements.

5. EQ-5D-5L (see: https://euroqol.org/eq-5d-instruments/eq-5d-5l-about/)

6. EQ-VAS (see: https://euroqol.org/eq-5d-instruments/eq-5d-5l-about/)

7. Do you have any other comments?

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Thank you very much for completing the questionnaire!

Appendix E: Factor loadings of participants

Participant ID	Factor 1	Factor 2	Factor 3	Factor 4
01	0.83*	0.04	-0.01	0.10
02	0.07	-0.24	0.61*	-0.17
03	-0.37	-0.07	0.05	-0.38
04	0.41	0.00	0.59*	-0.24
05	0.45*	0.32	0.14	0.11
06	-0.11	0.14	0.62*	0.17
07	0.16	0.56*	0.03	0.24
08	0.20	0.47*	-0.12	-0.01
09	0.16	0.44*	0.24	0.21
10	0.76*	-0.12	0.14	-0.11
11	-0.03	-0.26	0.75*	-0.02
12	-0.26	-0.08	0.18	0.63*
13	0.53*	-0.03	0.14	0.39
14	0.50*	-0.28	0.31	-0.06
15	-0.11	0.15	0.66*	0.16
16	0.63*	-0.19	0.03	0.30
17	0.53*	0.30	-0.13	0.03
18	0.04	0.00	0.41*	0.35
19	0.80*	-0.19	-0.14	0.04
20	0.60*	0.15	-0.06	-0.02
21	-0.26	0.72*	0.02	-0.10
22	0.58*	-0.12	-0.20	-0.12
23	0.47*	0.26	-0.17	0.29
24	0.68*	0.35	-0.10	-0.15
25	0.66*	-0.04	0.05	0.26
26	-0.31	0.21	0.60*	0.27
27	0.70*	0.05	0.07	-0.26
28	0.45*	0.00	0.29	0.27
29	0.29	0.06	0.01	0.70*
30	-0.16	-0.13	0.51	0.51
31	0.61*	0.10	0.06	-0.05
32	0.28	0.22	0.31	0.26
33	0.75*	0.18	0.15	0.07
34	0.50	0.42	0.33	0.02
35	0.27	0.35	0.44	-0.32

Participant ID	Factor 1	Factor 2	Factor 3	Factor 4
36	0.12	0.35	0.67*	-0.20
37	0.24	-0.29	0.43*	0.02
38	0.34	0.13	0.18	0.57*
39	0.06	0.08	0.66*	0.16
40	0.65*	-0.12	-0.09	-0.08
41	-0.34	-0.09	0.60*	0.29
42	0.54*	0.26	-0.18	0.33
43	0.34	0.08	0.60*	0.17
Eigen Value	9.1	2.8	5.7	3.1
Explained variance (%)	21.1	6.5	13.1	7.3
Exemplars(N)	19	4	12	3

*The participant 1) has a factor loading for the factor that is higher than the factor loading threshold with a p-value <0.05, and 2) has a square loading on the factor that is higher than their sum of squares loading on all other factors

4

To What Extent Do Citizens Support The Disinvestment Of Healthcare Interventions?

An exploration of the support for four viewpoints on active disinvestment in the Netherlands

Rotteveel AH
Lambooij MS
van Exel NJA
de Wit GA

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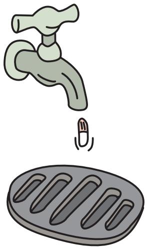

Abstract

Background: Active disinvestment of healthcare interventions (i.e. discontinuing reimbursement by means of a policy decision) has received limited public support in the past. Previous research has identified four viewpoints on active disinvestment among citizens in the Netherlands. However, it remained unclear how strong these viewpoints are supported by society, and by whom. Therefore, the current study aimed to 1) measure the support for these four viewpoints and 2) assess whether support is associated with background characteristics of citizens.

Method: In an online survey, a representative sample of adult citizens in the Netherlands (n=1,794) was asked to rate their agreement with short narratives of the four viewpoints on a 7-point Likert scale. The survey also included questions on sociodemographic characteristics, health status, healthcare utilization, and opinions about responsibility and costs in the healthcare context. Logistic regression models were estimated for each viewpoint to assess the association between viewpoint support and these characteristics.

Results: The support for the different viewpoints varied between 46.8% and 57.7% of the sample. Viewpoint support was associated with participants' age, gender, educational level, financial situation, healthcare utilization, opinion on the responsibility of the government for the health of citizens, and opinion on whether the increase in healthcare expenditure and health insurance premiums is considered a problem.

Conclusion: Resistance to active disinvestment may partially be explained by the consequences of disinvestment citizens anticipate experiencing themselves. Citizens considering the increase in healthcare expenditure a larger problem were more supportive of disinvestment than those considering it less of a problem.

Introduction

In many countries, healthcare expenditures have been increasing due to innovation, changes in the demand for healthcare and demographic developments [1, 2]. Healthcare expenditures have been rising faster than the gross domestic product of countries, putting pressure on the public financing of healthcare [1, 2]. As a result, the affordability of healthcare in the future is of major concern for policymakers. To keep healthcare affordable, policymakers have been taking different cost containment measures [3]. One of such measures is Health Technology Reassessment (HTR) [4, 5]. HTR is the structured, evidence-based reassessment of healthcare interventions currently being used and reimbursed, based on their clinical, economic, social and ethical aspects [4, 5]. In case HTR shows that healthcare interventions do not meet current requirements for reimbursement, one may argue that these interventions need to be actively disinvested. With active disinvestment we mean the partial or full withdrawal of reimbursement by means of a policy decision [6, 7].

Previous studies have estimated that, in OECD countries, about 20 to 30% of the healthcare budget is spent on ineffective healthcare interventions [8, 9]. Hence, a lot could possibly be gained from HTR and disinvestment, both in terms of health and healthcare expenditures. Nonetheless, HTR initiatives are scarce and disinvestment rarely takes place [10, 11]. Even in cases where there was clear evidence of ineffectiveness or unsafety, proceeding to disinvestment appeared to be difficult [5, 12]. Support among the public, policymakers and healthcare providers has been shown to be essential to proceed to actual disinvestment [6, 10]. Previous research showed that in cases where such support was limited, the disinvestment process was terminated at some point and reimbursement of the healthcare intervention concerned was maintained [10].

In a previous study from the Netherlands, four viewpoints on active disinvestment of healthcare interventions were identified among citizens, namely: 1) Maintain necessary healthcare, even if it is expensive or only results in small health gains, 2) Maintain necessary healthcare, if objectively determined and if there is no support for disinvestment, 3) Disinvest unnecessary, ineffective and inefficient healthcare, and 4) Transparent and consistent disinvestment decision-making processes [13]. Although the method used to elicit these viewpoints, Q-methodology [14], is very useful for obtaining rich, holistic descriptions of the different viewpoints present in society, it provides no information about the degree of support for these viewpoints in society, or how these are related to background characteristics of the citizens supporting them [15]. Knowing not only the societal viewpoints on active disinvestment, but also the degree of support for the viewpoints may be relevant if policymakers wish to be responsive to the preferences of citizens with regards to the disinvestment of healthcare interventions [16]. Insight in how widespread the support for the different viewpoints is, may provide policymakers with guidance on approaches to disinvestment (regarding decision-making and communication strategies) that may receive most public support. In addition, knowing how the support for the viewpoints relates to the background characteristics of citizens may provide insight in the factors explaining viewpoint support and guidance on targeted communication strategies on disinvestment.

The first aim of this study is to obtain insight into the support for the four previously identified viewpoints on active disinvestment of healthcare interventions among citizens in the Netherlands. The second aim is to assess whether the support for the different viewpoints was associated with background characteristics of citizens, i.e. their sociodemographic characteristics, health status, healthcare utilization, and opinions about responsibility and costs in the healthcare context.

Methods

Online survey

Data was collected using an online survey. To enable participants to assess their level of agreement with the four viewpoints, we asked participants to rate their agreement with short narratives for the four viewpoints, displayed in Table 1. We chose this approach over other approaches, such as rating or ranking a selection of the statements from the underlying Q-methodology study, as this approach enables participants to assess the different viewpoints in a holistic way [15, 16].

In the formulation of the narratives, we aimed to capture the key elements of the different viewpoints. For this reason, we focussed on the aspects that were identified as characterising and distinguishing for the viewpoints in the underlying study [13], as suggested by previous studies [15, 16]. The summaries were edited to make them clear and concise enough for assessment by citizens in a survey. In this editing process, we tried to ensure that each description covered the essence of the viewpoint and was sufficiently distinct from the narratives for the other viewpoints.

Table 1: Short narratives for the four viewpoints

#	Title	Narrative[a]
1	Maintain necessary healthcare, even if it is expensive or only results in small health gains	Treatments that are necessary must continue to be reimbursed. Necessary treatments are treatments for critically ill patients, treatments that are listed in the medical guidelines and treatments that doctors believe to be necessary. If a treatment exists, it is morally unacceptable to deny it to a patient. Even if treatment has little effect, is very expensive, or if the quality of life is still poor after treatment, the reimbursement may not be discontinued.
2	Maintain necessary healthcare, if objectively determined and if there is no support for disinvestment	Treatments that are necessary must continue to be reimbursed, regardless of the costs. However, it is important that necessity is established based on research (objective) rather than based on the patient's opinion or wishes. If the quality of life is still poor after treatment or if patients are able to pay for the treatment themselves, reimbursement of this treatment may be discontinued. However, this must be supported by society and must not happen at the expense of vulnerable groups.
3	Disinvest unnecessary, ineffective and inefficient healthcare	It is important that we are conscious of our healthcare expenditure. That is why discontinuing reimbursement is morally acceptable in certain situations. For example, the reimbursement of treatments that are not necessary, that do not work or that are expensive in relation to the health they provide must be discontinued. It is not important whether patients themselves think they benefit from a treatment. Treatments for diseases that are contagious or life-threatening to young people should continue to be reimbursed. Preventive care must also continue to be reimbursed.
4	Transparent and consistent disinvestment decision-making processes	Decisions about discontinuing reimbursement should always be made in the same way and properly explained. In this way, everyone is treated equally, and people understand the decision better. Treatments that work well, are necessary, or help patients to function better in society must continue to be reimbursed. However, we must make sure that health care remains affordable. That is why costs must be considered in decisions about discontinuing reimbursement.

[a] The narratives were originally in Dutch for the purpose of the survey and were translated into English language for this paper by a professional translation company

In the survey, participants were shown the four narratives in a randomized order, each on a separate page, and unlabelled (i.e. without the title as shown in Table 1). Participants were asked to express their agreement with the narratives using a 7-point Likert scale ranging from completely disagree (1) to completely agree (7). After having rated all four narratives, participants were presented with their rating of the narratives on an overview page, and asked to check and, if desired, adjust their ratings. Finally, we asked participants who rated all four narratives with a 4 or lower, indicating they did not agree with any of them, to describe their viewpoint on active disinvestment of healthcare interventions in their own words. With this question, we aimed to identify any important societal viewpoints that may have been missed in the underlying Q-methodology study.

To measure whether support for the viewpoints is associated with background characteristics of citizens, a number of questions were added to the survey (see Appendix A). These background characteristics were selected because previous research showed that these characteristics may be associated with citizens' views or preferences regarding healthcare priority setting [16-21]. The survey questions covered the sociodemographic characteristics age, gender, educational level, and financial situation of participants. Financial situation was measured by asking participants to what degree their household could make ends meet. The survey also included questions on health status and healthcare utilization. Health status was measured using the EQ-5D-5L Dutch version [22]. The questions on healthcare utilization concerned the number of times participants visited the general practitioner and hospital for themselves in the last year.

In addition, as we hypothesized that viewpoint support may be related to how people think about responsibility and costs in the healthcare context, we included four questions asking participants about their opinion on these topics: 1) To what extent do you think the government is responsible for **the health of Dutch people?**, 2) To what extent do you think the government is responsible for **healthcare in the Netherlands**?, 3) To what extent do you see **rising healthcare costs** as a problem?, and 4) To what extent do you see **rising health insurance premiums** as a problem? Participants were asked to rate their opinion on these topics on a 5-point Likert scale.

The survey was pilot tested in a group of 16 citizens, varying in age, gender and educational attainment, recruited by a commercial panel organization. Participants were asked for their feedback on whether the instructions and survey questions (including the viewpoint narratives) were comprehensible. The pilot test showed that the survey was sufficiently clear and feasible, and that no adaptations to the survey were necessary. As the survey was not changed between the pilot test and main study, the responses of the pilot test participants were retained in the final dataset.

Data collection

Data was collected from a representative sample of adult citizens in the Netherlands, based on quota-sampling by age, gender, educational level and region of residence. Participants were recruited by a commercial panel organization. Selected members of their panel received an invitation by e-mail, which contained information on the aim and the organization conducting the study, the content and length of the survey, and data management. If panel members accepted the invitation to participate, their informed consent was obtained at the start of the survey.

The research project has been assessed by the Centre for Clinical Expertise (CCE) at the National Institute for Public Health and the Environment (RIVM) in the Netherlands. The CCE concluded that the research project is exempted from further review by a medical ethics committee as it does not fulfil the specific conditions as stated in the Dutch Medical Research Involving Human Subjects Act.

Data analysis

To estimate the proportion of participants supporting each viewpoint, a dummy variable was defined for each viewpoint. The cut-off value for these dummy variables was set at 6 (i.e. agree) on the 7-point Likert scale, with participants rating the narrative with a 6 or higher being classified as supporting the particular viewpoint. A cut-off value of 6, thus excluding those who rated the narrative with a 5 (i.e., agree a little), was chosen in order to focus the analysis on participants who were fairly confident that they agreed with the narrative.

In order to also estimate the distribution of support for the viewpoints in society, we matched participants to the viewpoint they agreed with most strongly, on the condition that they supported this viewpoint (i.e. rated the corresponding narrative with a 6 or higher). Overall support for a viewpoint was calculated as the proportion of participants matched to that viewpoint. If a participant agreed most with multiple viewpoints (i.e. rated more than one narrative highest), they were split over these viewpoints (e.g. 50% viewpoint 3 and 50% viewpoint 4). If participants did not support any of the viewpoints (i.e. rated all narratives with a 4 or lower), they were categorized as ‘none’. Participants whose highest rating of any viewpoint was 5 were categorized as ‘moderate’ in order to distinguish them from the ‘none’ group, as they still elicited some support for at least one of the viewpoints.

To determine whether and how background characteristics of citizens were associated with support for the viewpoints, we estimated four logistic regression models, one for each viewpoint. The dependent variable was support for the respective viewpoint, as explained in the first paragraph of this section. In model formulation, a stepwise approach was taken. First, a model with the sociodemographic, health status and healthcare utilization characteristics as independent variables was estimated. Second, the opinion variables were added to the model to inspect whether they improved the models in terms of the Akaike's Information Coefficient (AIC). All analyses were conducted in R, version 4.0.2 [23].

Results

Data was collected in November and December 2020. In total, 4,446 panel members were invited to participate in this study, of whom 2,283 (51.3%) clicked on the link to the survey. Of these potential participants, 2,182 (95.6%) provided informed consent and 1,818 (79.6%) also completed the survey. Of these, 17 participants were excluded because they took more than 20 minutes to complete one of the four viewpoint questions, indicating that they may not have had their full attention with the survey. Furthermore, 7 participants were excluded because they gave the four descriptions all the same score (i.e. straight lining) and had a response time to the viewpoint questions in the 5th fastest percentile (i.e. speeding). This left 1,794 participants for the analysis.

Table 2 shows the descriptive statistics of the participants, and, for the sampling variables, the reference values in the general population of the Netherlands. Table 2 indicates that our sample was representative with regards to gender, educational level and region of residence, as intended. With regards to age, participants aged 66 to 75 years were overrepresented, while participants younger than 26 years and participants older than 75 years were slightly underrepresented.

Table 2: Descriptive statistics of the sample and the general population of the Netherlands

		Sample (n=1,794)		General population[a]
		N (%)	Mean (SD[b])	%
Sampling characteristics				
Age			50.8 (18.0)	
	18 - 25 years	190 (10.6)		12.5
	26 - 35 years	254 (14.2)		15.8
	36 - 45 years	258 (14.4)		14.7
	46 - 55 years	297 (16.6)		17.8
	56 - 65 years	325 (18.1)		16.4
	66 - 75 years	331 (18.5)		13.3
	> 75 years	139 (7.7)		9.5
Gender	Male	905 (50.4)		49.7
	Female	886 (49.4)		50.4
	Other	3 (0.2)		-
Educational level[c]	Low	578 (32.2)		30.2
	Middle	642 (35.8)		36.8
	High	574 (32.0)		31.5
Region (province) of residence	Drenthe	66 (3.7)		2.8
	Flevoland	47 (2.6)		2.4
	Friesland	52 (2.9)		3.7
	Gelderland	204 (11.4)		12.0
	Groningen	65 (3.6)		3.4
	Limburg	118 (6.6)		6.5
	Noord-Brabant	259 (14.4)		14.7
	Noord-Holland	269 (15.0)		16.5
	Overijssel	101 (5.6)		6.7
	Utrecht	125 (7.0)		7.8
	Zeeland	41 (2.3)		2.2
	Zuid-Holland	440 (24.5)		21.3
	Missing	7 (0.4)		-
(Other) Sociodemographic characteristics				
Financial situation of household	We're not making ends meet at all	33 (1.8)		
	We're not quite making ends meet	90 (5.0)		
	We can make ends meet	440 (24.5)		
	We can comfortably make ends meet	832 (46.4)		
	We are financially very comfortable	362 (20.2)		
	Rather not say	37 (2.1)		
Health and healthcare utilization				
Health status	EQ-5D-5L utility score[d]		0.862 (0.174)	

Table 2 continued:

		Sample (n=1,794)		**General population**[a]
		N (%)	Mean (SD[b])	%
Visited General Practitioner (GP) last year	Not at all	536 (29.9)		
	1 or 2 times	906 (50.5)		
	3 to 5 times	255 (14.2)		
	More than 5 times	93 (5.2)		
	Rather not say	4 (0.2)		
Visited hospital last year	Not at all	992 (55.3)		
	1 or 2 times	526 (29.3)		
	3 to 5 times	170 (9.5)		
	More than 5 times	102 (5.7)		
	Rather not say	4 (0.2)		
Opinion				
To what extent do you think the government is responsible for **the health of Dutch people**?	Not responsible	87 (4.8)		
	A little responsible	360 (20.1)		
	Moderately responsible	466 (26.0)		
	Responsible	683 (38.1)		
	Very responsible	198 (11.0)		
To what extent do you think the government is responsible for **healthcare in the Netherlands**?	Not responsible	21 (1.2)		
	A little responsible	120 (6.7)		
	Moderately responsible	117 (6.5)		
	Responsible	829 (46.2)		
	Very responsible	707 (39.4)		
To what extent do you see **rising healthcare costs** as a problem?	No problem at all	25 (1.4)		
	A small problem	116 (6.5)		
	A moderate problem	518 (28.9)		
	A large problem	867 (48.3)		
	A very large problem	268 (14.9)		
To what extent do you see **rising health insurance premiums** as a problem?	No problem at all	21 (1.2)		
	A small problem	125 (7.0)		
	A moderate problem	513 (28.6)		
	A large problem	760 (42.4)		
	A very large problem	375 (20.9)		

[a] Source: CBS Statline: https://opendata.cbs.nl/statline/#/CBS/en/

[b] SD: standard deviation

[c] Educational levels correspond to the SOI 2016 and the ISCED 2011 classifications

[d] Calculated from the EQ-5D-5L score based on the Dutch tariff [22], using the eq5d package for R [24]

Support for the four viewpoints

The rating participants gave to the four viewpoint narratives is displayed in Figure 1. Viewpoint 1, which most strongly opposes disinvestment was rated most often with scores indicating disagreement. However, this viewpoint also most often received the score of 7 (completely agree), indicating a stronger preference both on the positive and on the negative side for this viewpoint, compared to the other viewpoints.

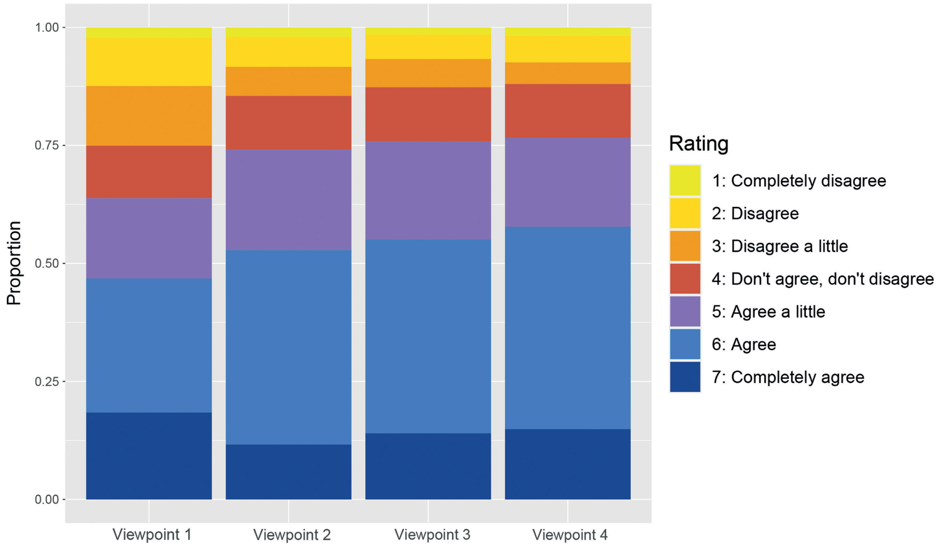

Figure 1: Stacked bar chart of the rating of the viewpoints by participants

Table 3 displays the descriptive statistics of the viewpoint ratings and support for the viewpoints. It shows that viewpoint 1 is supported by a small minority of 46.8% of the sample. The other viewpoints are supported by a small majority of the sample, with viewpoint 4, which is about a transparent and consistent disinvestment processes, receiving most support (i.e. 57.7%).

Table 3: Descriptive statistics and support for the viewpoints

				Correlation			Support[a]	
	Viewpoint	**Mean**	**SD**	**V2**	**V3**	**V4**	**N**	**%**
1	Maintain necessary healthcare, even if it is expensive or only results in small health gains	4.9	1.7	-0.09	-0.17	-0.08	840	46.8
2	Maintain necessary healthcare, if objectively determined and if there is no support for disinvestment	5.1	1.5		0.33	0.27	948	52.8
3	Disinvest unnecessary, ineffective and inefficient healthcare	5.2	1.4			0.28	989	55.1
4	Transparent and consistent disinvestment decision-making processes	5.3	1.4				1,036	57.7

[a] Support was determined based on a cut-off point of 6: Participants rating the viewpoint 6 or higher on the 7-point Likert scale were classified as supporting the viewpoint.

Figure 2 shows the result of the matching of participants to the four viewpoints and the categories none (i.e. supporting none of the viewpoints) and moderate (i.e. moderately supporting the viewpoints). The data underlying this matching is displayed in Appendix B, which shows that 40% of participants was matched to a single viewpoint, 28% was split over 2 viewpoints, 17% was split over 3 viewpoints and 6% was split over 4 viewpoints. Despite of this overlap in viewpoint support within participants, Table 3 shows that the correlations between the viewpoints is low to moderate, indicating that these can still be interpreted as distinct viewpoints. Figure 2 shows that the smallest proportion of participants (19.4%) was matched to viewpoint 2 and the largest proportion of participants (25.6%) was matched to viewpoint 1. Furthermore, Figure 2 shows that 8.9% of participants was not matched to a viewpoint, with 6.9% being classified as moderate and 2.0% being classified as supporting none of the viewpoints. This very low proportion of participants supporting none of the viewpoints indicates that it is likely that no important viewpoints were missed in the underlying Q-methodology study. Of the 36 participants supporting none of the viewpoints, 19 (53%) provided an informative description of their viewpoint on active disinvestment. Nine of them were generally against any form of disinvestment, five felt that treatment decisions can only be taken on an individual basis, two felt they had insufficient information, two stated that disinvestment is not necessary as public money could be saved elsewhere (e.g. in other public sectors), and one had the opinion that only cosmetic interventions should be disinvested.

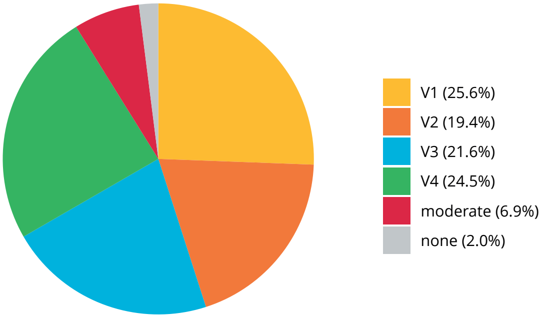

Figure 2: Proportion of participants supporting the viewpoints

Variables associated with viewpoint support

Table 4 presents four logistic regression models that were estimated to assess whether the support for the four viewpoints was associated with the sociodemographic characteristics, health, and healthcare utilization of citizens. Because of missing values on independent variables, 45 participants (3%) were excluded from the regression analyses. This left 1,749 participants for the estimation of the logistic regression models.

The results in Table 4 show that older participants were more likely to support viewpoints 2, 3, and 4. Furthermore, females were less likely to support viewpoints 2 and 3 than males. Higher educated participants were less likely to support viewpoint 1. With regards to financial situation, participants in a more financially comfortable position were less likely to support viewpoint 1 and more likely to support viewpoints 3 and 4. The health status of participants was not associated with viewpoint support. Participants who had visited the GP last year were less likely to support viewpoint 3, the viewpoint that is most open to disinvestment, while people who had visited the hospital last year were more likely to support viewpoints 1 and 2, which by and large oppose disinvestment.

Table 4: Model results of the logistic regression models estimating the association between viewpoint support and the sociodemographic, health status and healthcare utilization variables

		V1[a]	V2[b]	V3[c]	V4[d]
Variable		**Coefficient**	**Coefficient**	**Coefficient**	**Coefficient**
Intercept		0.89***	-0.12	-0.05	-0.42
Age		-0.01	0.01*	0.01*	0.01*
Gender (ref=Male)	Female	-0.07	-0.36***	-0.37***	-0.11
Education level (ref=Low)	Middle	-0.34*	0.04	-0.27*	0.15
	High	-0.90***	0.21	0.12	0.23
Financial situation household (ref= We can make ends meet)[e]	We're not making ends meet	0.03	-0.29	-0.29	-0.21
	We can comfortably make ends meet	-0.27*	-0.18	0.27*	0.29*
	We are financially very comfortable	-0.54***	-0.10	0.53***	0.50***
Health status (ref=Worse)[f]	Better	-0.17	0.05	0.08	0.20
Visited GP last year (ref= Not visited GP)[g]	Visited GP at least once	0.15	-0.06	-0.23*	0.01
Visited hospital last year (ref= Not visited hospital)[h]	Visited hospital at least once	0.35**	0.24*	0.16	-0.14
AIC		2,328	2,402	2,350	2,354

[a] Viewpoint 1: Maintain necessary healthcare, even if it is expensive or only results in small health gains

[b] Viewpoint 2: Maintain necessary healthcare, if objectively determined and if there is no support for disinvestment

[c] Viewpoint 3: Disinvest unnecessary, ineffective and inefficient healthcare

[d] Viewpoint 4: Transparent and consistent disinvestment decision-making processes

[e] As the lower two categories of this variable included a very small number of participants, these categories were combined into the category 'We're not making ends meet'. As this category was still quite small, the category 'We can make ends meet' was chosen as the reference category.

[f] As the data of this variable was skewed, participants were divided into two categories with the median EQ-5D value in the sample as the cut-off point.

[g] As the number of people that visited the GP multiple times last year was small, this variable was dichotomized, with 'not visited GP' being compared to 'visited GP at least once'.

[h] As the number of people that visited the hospital multiple times last year was small, this variable was dichotomized, with 'not visited hospital' being compared to 'visited hospital at least once'

* P-value ≤0.05; ** P-value ≤0.01; *** P-value ≤0.001

Table 5 shows the results of additional analyses to assess whether adding the opinion variables improved the regression models shown in Table 4. As participants' opinion on the extent to which the Dutch government is responsible for healthcare did not add to any of the four models (i.e. AIC increased after adding this variable), this variable was not included in the models displayed in Table 5. The other three variables were retained in the models displayed in Table 5 as the AIC of at least one of the four models decreased when adding these variables. Table 5 shows that participants who considered the increase in healthcare expenditure a large problem were more likely to support viewpoints 2, 3 and 4, and less likely to support viewpoint 1. Furthermore, participants who considered the increase in health insurance premiums a (very) large problem were more likely to support viewpoints 1 and 4. Finally, participants who considered the government more responsible for the health of Dutch people were more likely to support viewpoint 1.

Table 5: Model results of the logistic regression models estimating the association between viewpoint support and the sociodemographic, health status, healthcare utilization and opinion variables

		V1[a]	V2[b]	V3[c]	V4[d]
Variable		**Coefficient**	**Coefficient**	**Coefficient**	**Coefficient**
Intercept		0.66*	-0.13	-0.44	-0.35
Age		-0.01**	0.01*	0.01*	0.01*
Gender (ref=male)	Female	-0.02	-0.39***	-0.40***	-0.17
Education level (ref=low)	Middle	-0.32*	0.02	-0.29*	0.13
	High	-0.84***	0.18	0.10	0.17
Financial situation household (ref= We can make ends meet)	We're not making ends meet	-0.07	-0.26	-0.28	-0.22
	We can comfortably make ends meet	-0.19	-0.21	0.27*	0.24
	We are financially very comfortable	-0.39*	-0.16	0.50**	0.38*
Health status (ref=worse)	Better	-0.14	0.03	0.06	0.18
Visited GP last year (ref= Not visited GP)	Visited GP at least once	0.13	-0.06	-0.23*	0.02
Visited hospital last year (ref= Not visited hospital)	Visited hospital at least once	0.30**	0.26*	0.17	-0.12
To what extent do you think the government is responsible for the health of Dutch people? (ref= Moderately responsible)[e]	Not / A little responsible	-0.06	-0.03	0.20	0.02
	Responsible	0.50***	-0.08	0.13	-0.13
	Very responsible	0.59**	-0.29	0.04	-0.33

Table 5 continued:

		V1[a]	V2[b]	V3[c]	V4[d]
Variable		**Coefficient**	**Coefficient**	**Coefficient**	**Coefficient**
To what extent do you see rising healthcare costs as a problem? (ref= A moderate problem)[f]	No / A small problem	0.57*	-0.12	-0.10	-0.31
	A large problem	-0.47***	0.33*	0.40**	0.73***
	A very large problem	-0.68**	0.40*	0.59**	0.54**
To what extent do you see rising health insurance premiums as a problem? (ref= A moderate problem)[g]	No / A small problem	-0.34	0.12	0.38	0.08
	A large problem	0.17	-0.10	0.08	-0.58***
	A very large problem	0.67***	-0.23	-0.23	-0.32
AIC		2,277	2,407	2,346	2,321

[a] Viewpoint 1: Maintain necessary healthcare, even if it is expensive or only results in small health gains

[b] Viewpoint 2: Maintain necessary healthcare, if objectively determined and if there is no support for disinvestment

[c] Viewpoint 3: Disinvest unnecessary, ineffective and inefficient healthcare

[d] Viewpoint 4: Transparent and consistent disinvestment decision-making processes

[e] As the lower two categories of this variable (i.e. not and a little) included a small number of participants these categories were combined into the category 'Not/a little responsible' and the moderate category was taken as the reference category

[f] As the lower two categories of this variable (i.e. no and a small problem) included a small number of participants these categories were combined into the category 'No/a small problem' and the moderate category was taken as the reference category

[g] As the lower two categories of this variable (i.e. no and a small problem) included a small number of participants these categories were combined into the category 'No/a small problem' and the moderate category was taken as the reference category

* P-value ≤0.05; ** P-value ≤0.01; *** P-value ≤0.001

Discussion

Support for the four viewpoints

The first aim of this study was to measure the public support in the Netherlands for four viewpoints on active disinvestment of healthcare interventions that were previously identified in the literature. We found that viewpoint 1 (maintain necessary healthcare, even if it is expensive or only results in small health gains) was supported by 46.8% of the sample, viewpoint 2 (maintain necessary healthcare, if objectively determined and if there is no support for disinvestment) by 52.8%, viewpoint 3 (disinvest unnecessary, ineffective and inefficient healthcare) by 55.1%, and viewpoint 4 (transparent and consistent disinvestment decision-making processes) by 57.7%. This indicates that if policymakers would be responsive to any one of these viewpoints and adapt the disinvestment process in such a way that it is completely in line with this viewpoint, they would likely receive support from approximately half of the citizens (depending on which viewpoint they choose), indicating that resistance of a considerable part of the population should be anticipated as well.

Furthermore, we found that many people supported multiple viewpoints to the same extent. This indicates that there are certain elements in the different viewpoints that people can agree with simultaneously. The previous Q-methodology study already identified one aspect that people supporting the different viewpoints all considered to be relevant in the context of disinvestment, namely transparency. Although people supporting viewpoint 4 agreed most strongly that transparency is important, it was also considered relevant by people supporting the other viewpoints [13]. Furthermore, Appendix B shows that many people support viewpoint 4 to the same extent as other viewpoints, further suggesting that transparency, but also consistency, are strongly supported in this context. Therefore, a policy option to increase public support for disinvestment is to ensure that such decisions are made in a consistent and transparent manner, both with regards to the underlying process as the rationale underlying these decisions.

To identify additional policy options to increase public support for active disinvestment, it may be interesting to further explore the common elements of the viewpoints. To this end, future research may consider organizing citizen councils or Delphi studies with citizens supporting the different viewpoints to explore whether consensus can be achieved on certain types of disinvestment policies. Another possibility could be to conduct preference studies for disinvestment policies to assess the differences and similarities in preferences between those supporting the different viewpoints.

Variables associated with viewpoint support

The second aim of this study was to assess whether support for the four viewpoints was associated with the sociodemographic characteristics, health and healthcare utilization and opinion of citizens. As in previous studies [16-20], we found that viewpoint support was associated with the sociodemographic characteristics age, gender and educational level. Furthermore, we found that these associations differed between the viewpoints (e.g. age was associated with support for viewpoint 2, 3 and 4, but not with support for viewpoint 1). In addition, we found that people in a better financial situation are less likely to support viewpoint 1 and more likely to support viewpoints 3 and 4, i.e. the viewpoints that are more favourable towards disinvestment. A possible explanation for this is that people who are financially comfortable may be more confident that if a treatment is disinvested, they will be able to finance the treatment themselves when they feel they need it. People who have trouble making ends meet, may worry more about future healthcare costs, causing them to be less supportive of viewpoints that are more favourable towards disinvestment.

With regards to the health and healthcare utilization variables, different from what we anticipated, health status was not associated with viewpoint support. This indicates that participants with a worse health status, who may anticipate a higher personal need for healthcare in the future, do not have a different viewpoint on disinvestment than those with a better health status. On the other hand, we did find that people who had visited the hospital last year, are more likely to support viewpoints 1 and 2, which are most opposed to disinvestment. Furthermore, people who had visited the general practitioner last year are less likely to hold viewpoint 3, which is most supportive of disinvestment. Hence, people potentially anticipating a higher need for healthcare in the future because of their current healthcare utilization are more reluctant to disinvestment compared to those potentially anticipating a smaller need for healthcare in the future. In line with the endowment effect [25], we hypothesize that a potential explanation for this is that people currently utilizing healthcare may be more worried that disinvestment will result in their current treatment or the follow-up treatments they anticipate to require in the future no longer being available to them.

With regards to the opinion variables, concerns about the increase in healthcare expenditures particularly were relevant in relation to viewpoint support. People who consider this a larger problem are more likely to support viewpoints 2, 3 and 4, and less likely to support viewpoint 1. This is not surprising given that viewpoint 1 reflects being reluctant to disinvestment. Hence, the reluctance to disinvestment may partially be explained by not considering the current increase in healthcare expenditures problematic.

People who consider the increase in health insurance premiums a very large problem are more likely to support viewpoint 1. This seems to contradict our finding that people considering the increase in healthcare expenditures a large problem are less likely to support viewpoint 1. A possible explanation for this seemingly contradictory finding may be that people may not be aware that healthcare expenditures and health insurance premiums are directly associated with each other, as was previously observed by Reckers-Droog et al. [26]. It may also be that participants assess these questions from a different perspective. For instance, participants may consider the increase in health insurance premiums problematic for themselves, while an increase in healthcare expenditures may be considered only relevant for the government or society at large, less for themselves.

To address these issues, policymakers may be recommended to better involve citizens in the complexity and necessity of disinvestment decisions. In addition to explaining citizens their choices and the need for disinvestment, policymakers may consider using public participation methods such as participatory value evaluation [27, 28] or citizen councils [29] to better involve citizens in disinvestment decision-making.

Validity of the four viewpoints

In this study, we found that only a small proportion of participants (2.0%) did not support any of the viewpoints. This proportion of participants not supporting any viewpoint was similar to previous studies estimating the support for viewpoints on healthcare priority setting in Europe [16, 30]. Combined with our finding that the descriptions of their viewpoint provided by these participants indicate that it is unlikely that an important viewpoint on active disinvestment was missed, the results of our current study appear to validate the findings of the underlying Q-methodology study.

There was a period of 1.5 years between the former Q-methodology study, identifying the viewpoints (in June & July 2019) and the current study, measuring the support for these viewpoints (in November & December 2020). In the meantime, Covid-19 spread across the world, resulting in a pandemic. In the Netherlands, as in many countries, this pandemic has put a considerable pressure on the healthcare system. Regular care was delayed or displaced [31] and healthcare providers worked extra hours to ensure that as many Covid-19 and other patients could be treated as possible. One could expect that such an extreme shock to the healthcare system may have affected the viewpoints of citizens on active disinvestment. For instance, as the pandemic has demonstrated the scarcity in healthcare resources, citizens may be more aware that choices must be made, making them more supportive of disinvestment. From our study, however, we can infer that no new viewpoints seem to have emerged during the pandemic and that societal viewpoints on disinvestment, therefore, appear to be quite stable despite of this pandemic. However, we cannot exclude a shift in the support for the four viewpoints as we have not measured viewpoint support before the pandemic. For instance, because of the demonstrated scarcity in healthcare resources, there may be more support for viewpoint 3 now, compared to the period before the Covid-19 pandemic. On the other hand, delays in access to healthcare may have made citizens more reluctant to disinvestment, increasing the support for viewpoint 1.

Strengths and limitations

The survey was administered to a large sample of citizens representative of the adult Dutch population with regards to gender, educational level and region of residence. Therefore, the findings of this study are expected to be generalizable to the population in the Netherlands to a large extent. In addition, we gave participants the opportunity to assess the four viewpoints in a holistic way by providing them with short narratives of the viewpoints rather than a number of separate statements. This enabled participants to make a well-considered evaluation of their agreement with these viewpoints. However, this study also has some limitations. First, people aged 66-75 years were overrepresented in this study. Considering that this age group is somewhat more likely to have used healthcare in the past year than younger age groups, and that healthcare utilization was associated with viewpoint support, the overrepresentation of this age group may have slightly affected the observed support for the different viewpoints.

Second, we found that many people supported multiple viewpoints simultaneously. Although this is not necessarily problematic as people can support multiple viewpoints simultaneously, it may also indicate that the narratives of the viewpoints were not sufficiently comprehensive or distinguishing. Although we formulated these narratives with four different authors (i.e. triangulation), considered the raw data of the underlying Q-methodology study in the formulation the narratives and pilot-tested them, we cannot exclude that some distinguishing characteristics may have been overlooked or that some narratives were not comprehensive enough. Furthermore, we can also not exclude that, despite of our efforts to explain disinvestment in the survey, some participants scored the narratives similarly because of unfamiliarity or overall disinterest with the topic of disinvestment.

Finally, this study was conducted in the Netherlands. Viewpoints and the support for them may be context specific. For instance, a study measuring the support for five viewpoints on healthcare priority setting across nine European countries, found that support for the viewpoints differed between countries [16]. Therefore, readers are recommended to take the specific context of this current study into account when transferring the results to a different context.

Conclusion

The four viewpoints on active disinvestment were each supported by approximately half of the participants. Many participants expressed support for multiple viewpoints, indicating that the narratives contained elements people can agree on simultaneously. Resistance to active disinvestment may be partially explained by the consequences citizens anticipate to experience from disinvestment themselves, either as a result of their (lack of) opportunity to finance disinvested healthcare themselves or their anticipated need for healthcare in the future. People who consider the increase in healthcare expenditures a large problem were more supportive of disinvestment than those considering it less of a problem.

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Appendices

Appendix A – Online survey

Next, there are a number of general questions. We use this information to determine whether opinions about the reimbursement of healthcare differ between people with different characteristics.
What is your age?
What is your gender?
What is the highest level of education or training you have completed?
What is the financial situation of your household at the moment?
What are the four digits of your postcode?

In the Netherlands, healthcare costs 5,500 euros on average per inhabitant per year. These costs are still rising every year. This is why, to keep care accessible and affordable for everyone, policymakers are critically examining whether new medical treatments should be included in the basic health insurance package. In the past, this was examined less critically. This could be why there are treatments in the basic benefit package that we do not want to reimburse at all anymore based on current knowledge. It is therefore important to also see whether treatments can be removed from the basic benefit package. Discontinuing reimbursement can create scope for reimbursing other, new treatments without increasing the cost of care. In this study, we examine what considerations citizens believe the government should take into account when making decisions about discontinuing the reimbursement of certain healthcare. We will shortly show you four descriptions of opinions about what the government should take into account when making decisions about discontinuing the reimbursement of certain forms of care. These opinions were taken from an earlier survey among citizens in the Netherlands. We would like to know to what extent you agree with these four opinions.	
Treatments that are necessary must continue to be reimbursed. Necessary treatments are treatments for critically ill patients, treatments that are listed in the medical guidelines and treatments that doctors believe to be necessary. If a treatment exists, it is morally unacceptable to deny it to a patient. Even if treatment has little effect, is very expensive, or if the quality of life is still poor after treatment, the reimbursement may not be discontinued.	- Completely agree - Agree - Agree a little - Don’t agree, don’t disagree - Disagree a little - Disagree - Completely disagree
Treatments that are necessary must continue to be reimbursed, regardless of the costs. However, it is important that necessity is established based on research (objective) rather than based on the patient’s opinion or wishes. If the quality of life is still poor after treatment or if patients are able to pay for the treatment themselves, reimbursement of this treatment may be discontinued. However, this must be supported by society and must not happen at the expense of vulnerable groups.	- Completely agree - Agree - Agree a little - Don’t agree, don’t disagree - Disagree a little - Disagree - Completely disagree

It is important that we are conscious of our healthcare expenditure. That is why discontinuing reimbursement is morally acceptable in certain situations. For example, the reimbursement of treatments that are not necessary, that do not work or that are expensive in relation to the health they provide must be discontinued. It is not important whether patients themselves think they benefit from a treatment. Treatments for diseases that are contagious or life-threatening to young people should continue to be reimbursed. Preventive care must also continue to be reimbursed.	- Completely agree - Agree - Agree a little - Don’t agree, don’t disagree - Disagree a little - Disagree - Completely disagree
Decisions about discontinuing reimbursement should always be made in the same way and properly explained. In this way, everyone is treated equally and people understand the decision better. Treatments that work well, are necessary, or help patients to function better in society must continue to be reimbursed. However, we must make sure that health care remains affordable. That is why costs must be considered in decisions about discontinuing reimbursement.	- Completely agree - Agree - Agree a little - Don’t agree, don’t disagree - Disagree a little - Disagree - Completely disagree
Below you can read the four opinions again. For each opinion, you will also see the extent to which you agree with that opinion. If you are satisfied with the answers you have given, please click on ‘continue’. If you want to change your answer, please do so here. Then click on ‘continue’.	
You have indicated that you do not agree with any of the opinions. Can you describe in a few sentences what your opinion is on the discontinuation of reimbursement?	

Below are a few questions about your health and your health care experience. We use this information to determine whether opinions about the reimbursement of care differ between people with a different health status and different experiences with health care.	
EQ-5D-5L Dutch version	
Have you seen a **GP for yourself in the past 12 months?** If so, how often?	- No - Yes, 1 or 2 times - Yes, 3 to 5 times - Yes, more than 5 times - Rather not say
Have you been to the **hospital for yourself in the past 12 months?** If so, how often?	- No - Yes, 1 or 2 times - Yes, 3 to 5 times - Yes, more than 5 times - Rather not say
To what extent do you think the government is responsible for **the health of Dutch people**?	- Not responsible - A little responsible - Moderately responsible - Responsible - Very responsible

To what extent do you think the government is responsible for **healthcare in the Netherlands**?	- Not responsible - A little responsible - Moderately responsible - Responsible - Very responsible
To what extent do you see **rising healthcare costs** as a problem?	- No problem at all - Small problem - Moderate problem - Large problem - Very large problem
To what extent do you see **rising health insurance premiums** as a problem?	- No problem at all - Small problem - Moderate problem - Large problem - Very large problem

Appendix B – Raw data underlying the matching of participants to viewpoints

Label	Matched to	N	%	%
V1	1	271	15.1%	40.0%
V2	2	122	6.8%	
V3	3	139	7.7%	
V4	4	185	10.3%	
Mixed on 2 views	1 and 2	66	3.7%	27.5%
	1 and 3	79	4.4%	
	1 and 4	80	4.5%	
	2 and 3	71	4.0%	
	2 and 4	80	4.5%	
	3 and 4	117	6.5%	
Mixed on 3 views	1, 2 and 3	49	2.7%	17.3%
	1, 2 and 4	49	2.7%	
	2, 3 and 4	171	9.5%	
	1, 3 and 4	41	2.3%	
Mixed on 4 views	all	115	6.4%	6.4%
Moderate	Moderate	123	6.9%	6.9%
None	None	36	2.0%	2.0%

5

Valuing Healthcare Goods And Services

A systematic review and meta-analysis on the WTA-WTP disparity

Rotteveel AH
Lambooij MS
Zuithoff NPA
van Exel J
Moons KGM
de Wit GA

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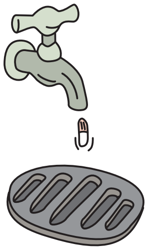

Abstract

Objective: To review the available evidence on the disparity between willingness-to-accept (WTA) and willingness-to-pay (WTP) for healthcare goods and services.

Methods: A tiered approach consisting of 1) a systematic review, 2) an aggregate data meta-analysis (AD-MA), and 3) an individual participant data meta-analysis (IPD-MA) was used. Medline, Embase, Scopus, Scisearch and Econlit were searched for articles reporting both WTA and WTP for healthcare goods and services. Individual participant data was requested from the authors of the included studies.

Results: Thirteen papers, reporting WTA and WTP from nineteen experiments/ subgroups, were included in the review. The WTA/WTP ratios reported in these papers, varied from 0.60 to 4.01, with means of 1.73 (median 1.31) for fifteen estimates of the mean and 1.58 (median 1.00) for nine estimates of the median. Individual data obtained from six papers, covering 71.2% of the subjects included in the review, yielded an unadjusted WTA/WTP ratio of 1.86 (95% CI: 1.52-2.28) and a WTA/WTP ratio adjusted for age, gender and income of 1.70 (95% CI: 1.42-2.02). Income category and age had a statistically significant effect on the WTA/WTP ratio. The approach to handling zero WTA and WTP values has a considerable impact on the WTA/WTP ratio found.

Conclusions and implications: The results of this study imply that losses in healthcare goods and services are valued differently from gains (ratio>1), but that the degree of disparity found depends on the method used to obtain the WTA/WTP ratio, including the approach to zero responses. Irrespective of the method used, the ratios found in our meta-analysis are smaller than the ratios found in previous meta-analyses.

Introduction

The healthcare market is characterized by many imperfections, such as asymmetric information between patients and physicians, third party payers, and uncertainty in demand and supply. Due to these market imperfections and government regulations, the price people pay for goods and services in the healthcare market does not necessarily reflect their value to them. Therefore, unlike the market for consumer goods, it is difficult to use revealed preferences to determine the value of healthcare goods and services [1]. To circumvent this problem, health economists have regularly resorted to using stated preferences methods, such as contingent valuation, to estimate the value of healthcare [2, 3].

An important application of stated preferences for healthcare is cost-benefit analysis [4-6]. In this context, two measures have been used for valuing healthcare: willingness-to-pay (WTP) and willingness-to-accept (WTA). WTP measures the amount of money one is willing to pay for obtaining a certain healthcare good or service. WTA measures the amount of monetary compensation one wants to receive for giving up a certain healthcare good or service. The relevant measure to use, thus, depends on the decision context, with WTP being used when people stand to gain something and WTA being used when people stand to lose something [4, 7, 8].

Previous studies have reported substantial differences between the WTP and WTA for the same good or service, both in hypothetical studies as well as in studies involving real transactions [9-11]. An aggregate data meta-analysis by Tunçel and Hammitt (2014) summarized the studies comparing WTP and WTA across different economic sectors. They reported an overall WTA/WTP ratio of 3.28, indicating that people, on average, want to receive a 3.28 times larger amount to give up a good or service than they are willing to pay to obtain this good or service. The size of the WTA/WTP ratio differed for the type of good valued, with studies on environmental goods reporting the largest WTA/WTP ratio of 6.23 on average [10]. A recent estimate of the WTA/WTP ratio for healthcare goods and services is lacking, indicating a knowledge gap on the WTA-WTP disparity for the healthcare sector. The only review reporting a separate WTA/WTP ratio for healthcare [12] dates back to 2002 and included only two studies reporting ratios of 1.9 and 6.4 [13, 14]. The more recent meta-analysis of Tunçel and Hammitt [10] did not look at healthcare separately but reported a mean ratio of 5.09 for health and safety goods together. Moreover, the search for this meta-analysis dates back to early 2012 and only covered one database (i.e. Econlit), indicating that the search could be updated and expanded to more databases to identify further relevant studies in the healthcare context.

In the literature, many different explanations for a disparity between WTA and WTP have been described. According to standard economic theory, WTA and WTP should be similar when the good valued is divisible and exchanged at zero transaction costs on an infinitely large market. If these conditions do not hold, WTA and WTP may be different. The size of this difference depends on income, the proportion of income that is spent on the good, and the income elasticity [5, 6]. Furthermore, the inability to substitute money for a (public) good, either because of perfect complementarity or because of asymptotic boundedness of the utility curve, may also be a reason for WTA to exceed WTP [15-17]. Moreover, according to several alternative economic theories, such as prospect theory, 1) people value a change from a reference point, instead of the final state after a change, and 2) the value function for losses is steeper than the value function for gains. For these reasons, WTA values are expected to be larger than WTP values [18-20].

It is important to obtain more insight in the WTA/WTP ratio for healthcare goods and services, as a disparity between WTA and WTP has important implications for healthcare decision-making, for example for reimbursement decision-making. If WTA is larger than WTP, a higher cost-effectiveness threshold may be used for decisions on stopping reimbursement of healthcare interventions as compared to decisions on starting reimbursement, in other words the cost-effectiveness ratio should probably be significantly less favourable for disinvestment to be welfare improving. In line with this, insight in the WTA/WTP ratio for healthcare goods and services may be helpful to better understand reimbursement decision-making as policy makers seem to find it more difficult to discontinue reimbursement than not to start reimbursement in the first place [21]. It may, therefore, also be important for researchers in the field of cost-benefit analysis of healthcare interventions and preference elicitation to obtain more insight in the WTA/WTP ratio for healthcare goods and services, as insight in this issue provides guidance on choosing the appropriate measure of the value of healthcare interventions given the decision context at hand, i.e. investment versus disinvestment of healthcare goods and services. Furthermore, insight in the WTA/WTP ratio for healthcare goods and services may be helpful in understanding the general reluctance of patients to change treatment despite potential advantages [22, 23], indicating that for a new treatment to be welfare improving it should offer substantially higher benefits to the patient than the current treatment.

The aim of this study is to review the available evidence on the disparity between WTA and WTP for healthcare goods and services in order to obtain an aggregated estimate of the WTA/WTP ratio for healthcare goods and services. To this end, we used a comprehensive tiered approach consisting of 1) a systematic review, 2) an aggregate data meta-analysis (AD-MA), and 3) an individual participant data meta-analysis (IPD-MA). Firstly, the systematic review provides an overview of published studies that compared WTP and WTA for healthcare goods and services. Secondly, the AD-MA, combines the estimates as reported in these studies. Finally, the IPD-MA enables us to calculate one overall estimate of the WTA-WTP disparity, to obtain more insight in the statistical and methodological uncertainty surrounding this estimate, and to correct the estimate for subject characteristics. The IPD-MA approach has not been applied before to estimate the WTA/WTP ratio. Hence, this study adds a new level of information to the previous literature.

Methods

Systematic review

The databases Medline, Embase, Scopus, Scisearch and Econlit were searched from inception to the search date (i.e. 9th or 13th of February 2017) using willingness-to-pay and willingness-to-accept (and variations thereof) in title, abstract or as keywords. For the databases that do not solely focus on health (i.e. Scopus, Scisearch and Econlit), the search strategy was extended with health-related search terms. The full search strategies are displayed in Appendix A.

After deduplication, titles and abstracts were screened for eligibility by two reviewers using the eligibility criteria in Table 1. If eligibility was not clear from title and abstract, the article was included in full-text screening to ensure that no eligible papers would be missed. Differences between reviewers were resolved by discussion. If consensus was not reached, a third reviewer was consulted. Full-text articles of all included abstracts were retrieved and screened for eligibility by one reviewer. If the reviewer was unsure about eligibility, the other reviewers were consulted.

Table 1: Eligibility criteria for the systematic review

Empirical studies (stated preferences)
Providing both willingness-to-pay and willingness-to-accept estimates: - for a comparable change - in healthcare goods or services - elicited in 1) the same subject or 2) two *randomly* allocated groups from the same sample
Published in English or Dutch
Full-text available

For each included article, the estimate of the WTA/WTP ratio was extracted. If several estimates for different subgroups or experiments were provided, all these estimates were extracted. Next to the WTA/WTP ratio, the following (study) characteristics were extracted: first author, year, country, good/service valued, number of study subjects (N), subject sample type, within versus between subject design, elicitation method, administration method, payment vehicle, and payment frequency (see Appendix B).

Aggregate data meta-analysis

From the WTA/WTP estimates extracted in the systematic review, an overall WTA/WTP ratio was calculated. This was calculated by taking the mean and median from the WTA/WTP estimates as reported by the studies. If studies only reported mean/median WTA and WTP at study-level (i.e. not a ratio), the WTA/WTP ratio at study-level was calculated by dividing WTA by WTP. Next to the mean and median, a weighted average WTA/WTP ratio was calculated to take account of large differences in number of subjects and number of estimates retrieved from studies [10, 11]. The estimates from the studies were weighted using this formula:

$$\frac{\sqrt{N_{ik}}}{\sqrt{K_i}}$$

where N is the sample size of estimate k from study i and K is the number of estimates provided by study i. As the aggregate WTA/WTP estimates were reported in different formats (i.e. mean, median or regression model estimate), overall WTA/WTP ratios were calculated for each format separately.

Individual participant data meta-analysis

Individual participant data (IPD) on WTP, WTA, age, gender and income was requested by sending an e-mail to the corresponding authors of the papers included in the AD-MA. If it was not possible to contact the corresponding author, other authors were e-mailed. If necessary, the authors were reminded twice. The retrieved IPD were analysed using three approaches increasing in complexity, which are described in the subsequent three paragraphs.

Descriptive analyses

Received datasets were merged and harmonized into one dataset for analysis. To facilitate comparison and analysis, all WTP and WTA values were converted to the same base year and currency unit (i.e. 2017 Euros, Dutch price level) using the OECD purchasing power parities [24] and the consumer price index from Statistics Netherlands [25]. To test whether the studies included in the IPD-MA were different from the studies included in the AD-MA an overall WTA/WTP ratio was calculated in a similar way to the AD-MA. To this end, study-level WTA/WTP ratios were calculated by dividing mean/median WTA at study-level by mean/median WTP at study-level. From these study-level WTA/WTP ratios, overall estimates were calculated by taking the mean, median and weighted average from these estimates.

Mixed model analysis

Of the 4213 subjects included in the IPD dataset, 302 subjects (7%) had a missing value on WTP, 218 subjects (5%) had a missing value on WTA, 1107 subjects (26%) had a missing value on both WTP and WTA, and 435 subjects (10%) had a missing value on income. As complete case analysis, i.e. exclusion of respondents with missing values, may introduce bias, multiple imputation of WTA, WTP and income was used. The imputation model used data on age, gender, income, country of study and converted WTA and WTP. We used a fully conditional specification with predictive mean matching to impute WTP and WTA when one was available and one was missing. The 1107 subjects with both WTP and WTA missing, were excluded because they missed both parameters of interest for this study. Data was imputed 10 times. All analyses were performed on each dataset separately and, subsequently, the results were pooled according to Rubin's rule [26]. As WTP, WTA and the WTA/WTP ratio were not normally distributed, the data was then log-transformed. As a result, respondents with WTA or WTP of zero were excluded from the analysis. As income was measured on different scales in different studies, income was dichotomized, based on median income (category) at study-level as a cut-off point. Subsequently, the log of WTA/WTP ratio was estimated with a linear mixed model. A random intercept was included to reflect any heterogeneity over studies in this outcome. The analysis was performed once without correction for covariates and once with correction for age, gender and income. All analyses were performed with SAS 9.4 software (SAS Institute Inc., Cary, NC, USA).

Sensitivity analyses: zero WTP and/or WTA

In the AD-MA and the descriptive analysis of IPD, data of subjects with zero WTP and/or WTA were included in the analysis. In the mixed model analysis of IPD, subjects with zero responses were excluded from the analysis because log-transformation of zero WTP and/or WTA is not possible. The best approach to dealing with zero responses in this context depends on the reasons behind zero responses (e.g. protest responses, not understanding the task or an actual very low/zero valuation [27-29]). In this meta-analysis, we were not able to determine the reason behind zero responses. Therefore, in order to assess the potential impact of our main approach to zero responses on the

WTA/WTP ratios, we conducted two sensitivity analyses. The first sensitivity analysis is the same as the descriptive analysis of IPD as described in descriptive analyses of the method section, however, excluding subjects reporting zero WTP and/or WTA. The second sensitivity analysis is the same as the mixed model analysis, described in the previous section, however, including subjects reporting zero WTP and/or WTA, by replacing their zero value by a ½, ⅓ or ¼ of the smallest value reported in the study concerned. This approach especially makes sense if subjects reported zero values because their WTP or WTA was too small to be picked up by the elicitation procedure used.

Results

Systematic review

Databases were searched on the 9th (Medline and Embase) and the 13th (Scopus, Scisearch and Econlit) of February 2017. In total, 396 records were identified of which, after removal of 231 duplicates, 165 remained for title and abstract screening. Of the 31 articles that were included in full-text screening, 13 were included in the review (see Appendix C). Figure 1 displays the PRISMA flow diagram.

Table 2 displays the descriptive characteristics and the extracted WTA/WTP ratios of the studies included in the systematic review. The 13 included studies provided estimates for 19 different experiments or subgroups.

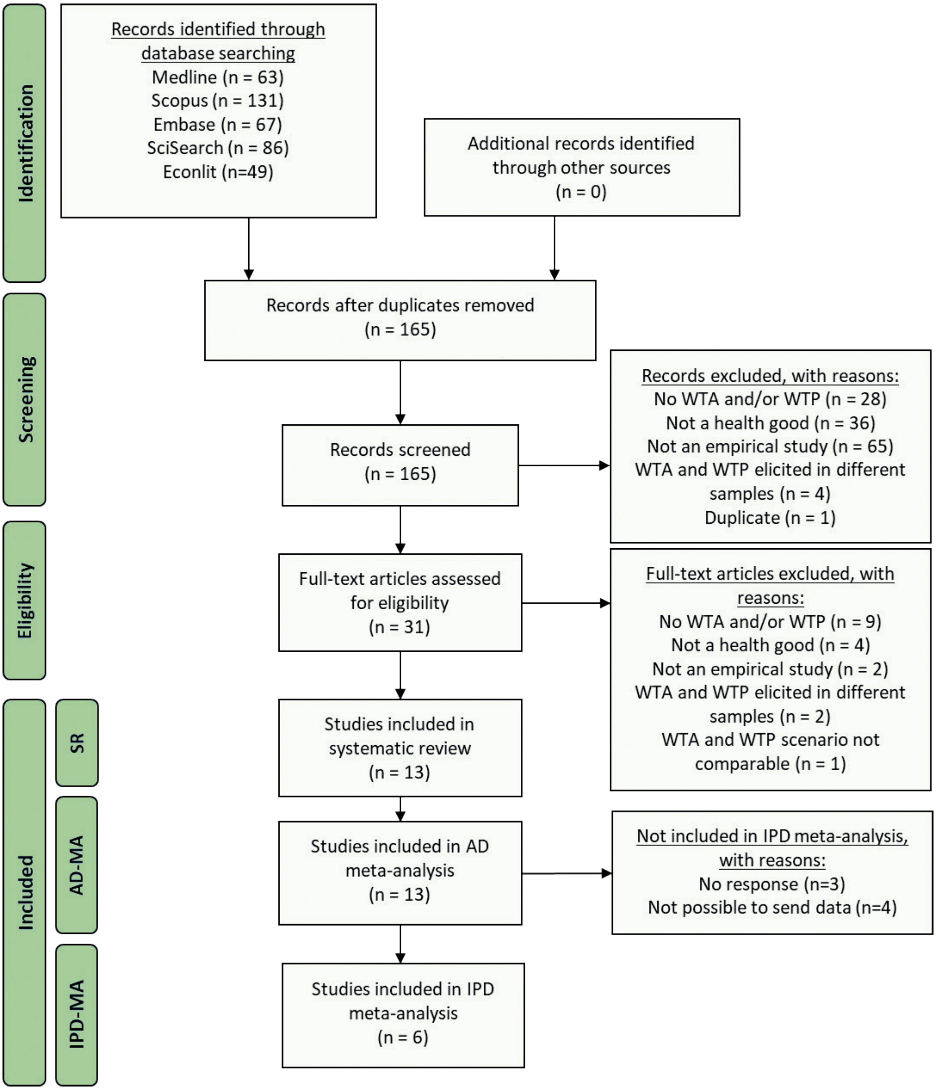

Figure 1: PRISMA flow diagram

Table 2: Descriptive characteristics and extracted WTA/WTP estimates of included studies

First author	Year	Country	Good/service valued	N	Subject sample type	Within/ between subject design	Elicitation method	Administration method	Payment vehicle; frequency	WTA/WTP ratio[a]		
										Mean	Median	Model estimate[b]
Bayen [30]	2016	France	Informal care	98	Suppliers	Within	Open-ended single question	Not clear	Not clear; hourly	0.70		
van den Berg[c] [31]	2005	The Netherlands	Informal care	149	Patients/ clients	Within	Close-ended question with open-ended follow-up	Surveys, not supervised	Tax (WTA) or out-of-pocket (WTP); weekly	1.05	1.00	
				149	Suppliers	Within	Close-ended question with open-ended follow-up	Surveys, not supervised	Tax (WTA) or out-of-pocket (WTP); weekly	1.22	1.00	
Borisova [32]	2003	United States	Methadone maintenance	303	Patients/ clients	Within	Open-ended single question	Surveys, supervised	Out-of-pocket; per visit	1.31		
Chiwaula [33]	2016	Malawi	Informal care	93	Suppliers	Within	Open-ended single question	Interviews	Out-of-pocket; once		2.40	
Finkelstein [34]	2016	Singapore	Life extending treatment at end of life	290	Patients/ clients	Within	Discrete choice experiment	Interviews	Public and out-of-pocket; once		0.77	
			Quality of life enhancing treatment at end of life	290	Patients/ clients	Within	Discrete choice experiment	Interviews	Public and out-of-pocket; once		0.77	
Grutters [35]	2008	The Netherlands	Hearing aid provision	291	Patients/ clients	Between	Discrete choice experiment	Interviews	Out-of-pocket; once			3.20
Manan[d] [36]	2015	Malaysia	Methadone maintenance	14	Patients/ clients	Within	Open-ended single question	Surveys, supervised	Out-of-pocket; per visit	0.60		
				15	Patients/ clients	Within	Open-ended single question	Surveys, supervised	Out-of-pocket; per visit	1.53		
				10	Patients/ clients	Within	Open-ended single question	Surveys, supervised	Out-of-pocket; per visit	0.81		
Martin-fernandez [37]	2010	Spain	Visit to family physician	404	Patients/ clients	Within	Payment card	Interviews	Not clear; not clear	3.30	1.55	

First author	Year	Country	Good/service valued	N	Subject sample type	Within/ between subject design	Elicitation method	Admin- istration method	Payment vehi- cle; frequency	WTA/WTP ratio[a]		
										Mean	Median	Model esti- mate[b]
Martin-fer- nandez [38]	2013	Spain	Primary care nursing consul- tation	662	Patients/ clients	Within	Payment card	(telephone) Interviews	Public (WTA) or out-of-pocket (WTP); once	1.45	2.00	
de Meijer [39]	2010	The Neth- erlands	Informal care	289	Patients/ clients	Within	Open-ended single question	Surveys, not super- vised	Public (WTA) or out-of-pocket (WTP); weekly	1.30		
				983	Suppliers	Within	Open-ended single question	Surveys, not super- vised	Public (WTA) or out-of-pocket (WTP); weekly	1.15		
O'Brien[e] [13]	1998	United States	Filgrastim (cancer drug)	107	General public	Within	Discrete choice experiment	Interviews	Insurance premium, monthly	2.38	NA[f]	
				109	General public	Within	Discrete choice experiment	Interviews	Insurance premium, monthly	1.61	3.75	
Tsuji [40]	2004	Japan	Telehealth	230	Patients/ clients	Within	Payment card	Surveys, not super- vised	Out-of-pocket, monthly	3.60	1.00	
Whynes [41]	2007	United Kingdom	Paediatric co- chlear implan- tation (hearing device)	64	Parents/ guard- ians	Within	Iterated close-end- ed question/ bidding game	Interviews	Public (NHS); monthly	4.01		

[a] Almost all estimates are ratios of mean/median WTA and WTP. Only Martin-Fernandez et al. (2010) reported mean and median of individual WTA/WTP ratios.

[b] Result estimated with a regression model for a person with median patient characteristics

[c] This study reported four estimates for two different experiments containing two subgroups each. The data of the subjects included in the second experiment (among informal caregivers and their care recipients), was also fully included in the study of de Meijer et al. (2010) and was, therefore, not extracted

[d] Experiments differed in household income of the subjects (i.e. low, medium and high)

[e] Experiments differed in the baseline cancer risk (i.e. 1/100 or 1/200 in the next 5 years). Median WTP for the first experiment is zero. Hence, the ratio could not be calculated.

[f] This ratio could not be calculated as median WTP is zero.

Aggregate data meta-analysis

The WTA/WTP ratios calculated from the extracted WTA/WTP estimates are displayed in Table 3. A mean WTA/WTP ratio of 1.73 for 15 mean estimates and of 1.58 for nine median estimates was found. The weighted average was 1.87 for mean estimates and 1.55 for median estimates. The small differences between the crude and weighted averages indicates that the estimates provided by studies with more subjects and/or more experiments/subgroups were not very different from other studies. One study [35] did not report mean or median but reported a regression model estimate of the WTA/WTP ratio instead. This estimate of 3.20 was relatively high compared to the mean WTA/WTP ratio for mean and median estimates.

Table 3: WTA/WTP estimates obtained from aggregate data

	Mean estimates	Median estimates	Regression model estimates
Mean	1.73	1.58	3.20
Weighted average	1.87	1.55	NA[a]
Median	1.31	1.00	3.20
Number of estimates (from n studies)	15 (10)	9 (7)	1 (1)

[a] NA: not applicable as it concerns one estimate

Individual participant data meta-analysis

From the 13 studies included in the AD-MA, six datasets were obtained for inclusion in the IPD-MA (see Figure 1). For the remaining seven studies, the data could not be included because of non-response (n=3) or because the authors were not able to send the data (n=4). The six datasets received covered 71.2% of the subjects that were included in the AD-MA, implying that the samples we could not include were relatively small compared to the samples we were able to obtain.

Descriptive analyses

Descriptive information of the six datasets received is displayed in Table 4. Of the 4,213 subjects included in the six datasets, 1,107 subjects were excluded from the analysis because they had both WTP and WTA missing. Of the remaining 3,106 subjects, 299 subjects (10%) reported a WTP of zero, 69 subjects (2%) a WTA of zero and 77 subjects (2%) both a WTP and a WTA of zero. This left 2,661 subjects for the mixed model analyses.

Table 5 displays the WTP and WTA per study after conversion to 2017 Euros (for raw data see Appendix D) and the results of the descriptive analysis. The study-level estimates of the WTA/WTP ratios were similar to the estimates found in the AD-MA. This indicates that the subsample of studies included in the IPD-MA was not that different from all studies included in the AD-MA. Appendix E shows the WTA/WTP ratio for different levels of age, gender and income. As expected, the ratio was higher in people with lower income compared to people with higher income. Furthermore, the two intermediate age groups reported lower WTA/WTP ratios compared to the youngest and the oldest age category.

Table 4: Descriptive information of the studies included in the IPD-MA

First author (year of publication)	Country	Currency (year of data collection)	Good/service valued	N[a]	Mean age	N males (%)	N low income[b] (%)	N subjects with missing values (%)		N subjects reporting zero WTP and/or zero WTA (%)		
								WTP missing	WTA missing	Only WTP is zero	Only WTA is zero	Both WTP and WTA are zero
van den Berg (2005) [31]	The Netherlands	Dutch guilders (2001)	Informal care	270	60.3	118 (44)	NA[c]	29 (11)	24 (9)	13 (5)	5 (2)	6 (2)
Borisova (2003) [32]	United States	US Dollars (1999)	Methadone maintenance	303	41.8	162 (53)	152 (50)	0 (0)	0 (0)	128 (42)	20 (7)	34 (11)
Chiwaula (2016) [33]	Malawi	Malawian Kwatcha (2013)	Informal care	92	41.9	65 (71)	NA[c]	0 (0)	0 (0)	0 (0)	0 (0)	1 (1)
Martin-Fernandez (2010) [37]	Spain	Euros (2008)	Visit to family physician	451	57.3	165 (37)	266 (59)	0 (0)	1 (<1)	30 (7)	12 (3)	4 (1)
Martin-Fernandez (2013) [38]	Spain	Euros (2011)	Primary care nursing consultation	653	65.2	255 (39)	341 (53)	0 (0)	5 (1)	70 (11)	2 (<1)	11 (2)
de Meijer (2010) [39]	The Netherlands	Euros (2002)	Informal care (one hour per week, suppliers)	992	54.3	289 (31)	491 (51)	239 (24)	143 (14)	29 (3)	21 (2)	12 (1)
			Informal care (one hour per day, patients)	345	67.1	141 (41)	103 (34)	34 (10)	45 (13)	29 (8)	9 (3)	9 (3)
Overall				3,106	57.2	1195 (39)	1,353 (51)	302 (10)	218 (7)	299 (10)	69 (2)	77 (2)

[a] This excludes subjects with both WTA and WTP missing

[b] As income was measured on different scales between studies (i.e. continuous versus categorical; different categories), income was dichotomized at study-level into low and high income with median income (category) at study-level as a cut-off point to facilitate the analysis. This column displays the proportion of subjects who were categorized into the low-income category. The remaining subjects were categorized as having a high income. [c] No income data was available for this study

Table 5: WTP, WTA and WTA/WTP estimates based on individual participant data (converted to 2017 Euros)

First author (year)	**WTP**			**WTA**			**WTA/WTP**[a]		
	N	Mean (SD[b])	Median (quartiles)	N	Mean (SD)	Median (quartiles)	N[c]	Mean	Median
van den Berg (2005) [31]	241	10.30 (5.85)	8.85 (5.90|11.80)	246	11.83 (7.19)	10.47 (8.85|14.75)	270	1.15	1.18
Borisova (2003) [32]	303	3.11 (6.23)	0.00 (0.00|3.78)	303	9.74 (13.60)	6.30 (1.10|12.60)	303	3.13	NA[d]
Chiwaula (2016) [33]	92	18.82 (23.80)	10.25 (6.15|20.50)	92	37.13 (36.60)	24.60 (14.35|48.69)	92	1.97	2.40
Martin-Fernandez (2010) [37]	451	26.09 (23.24)	23.94 (10.64|37.24)	450	47.94 (30.03)	47.22 (23.94|60.52)	451	1.84	1.97
Martin-Fernandez (2013) [38]	653	18.40 (19.35)	12.80 (6.40|23.60)	648	26.75 (21.92)	25.60 (12.80|38.40)	653	1.45	2.00
de Meijer (2010) [39][e]									
Suppliers	753	11.49 (7.30)	11.44 (8.58|14.29)	849	13.20 (8.51)	11.44 (8.58|14.29)	992	1.15	1.00
Patients	311	8.70 (6.73)	8.58 (5.72|11.44)	300	11.33 (8.17)	10.29 (5.72|14.29)	345	1.30	2.30
Mean							3,106	1.71	1.63
Weighted average							3,106	1.70	1.63
Median							3,106	1.45	1.59

[a] Calculated from study-level WTA and WTP: mean WTA/WTP= mean WTA/mean WTP; median WTA/WTP= median WTA/median WTP

[b] SD: standard deviation

[c] Subjects with either WTA or WTP missing were still included in this analysis. Therefore, this N is higher than the N for WTP and WTA separately.

[d] Median WTP is zero. Therefore, the WTA/WTP ratio based on medians could not be calculated for the study of Borisova et al.

[e] As informal caregivers reported WTP and WTA for one hour extra per week and patients reported WTP and WTA for one hour extra per day, the good valued is not comparable between these groups. Therefore, the subgroups are reported separately.

Mixed model analysis

Table 6 displays the results of the mixed model analysis. The unadjusted WTA/WTP ratio was 1.86 (95% confidence interval: 1.52-2.28). Age and income category both had a statistically significant effect on the WTA/WTP ratio found. The table in Appendix F displays the ln(WTA/WTP ratio) and the WTA/WTP ratio for different groups of subjects. The figures in Appendix F display the trend of the WTA/WTP ratio for different types of subjects, based on the ln(WTA/WTP) slope estimates. The largest difference in the WTA/WTP ratio of 0.45 was found between high-income 30-year olds and low-income 65-year olds. Furthermore, the difference between the low- and high-income groups increased with increasing WTA and WTP values.

Table 6: WTA/WTP ratios obtained from the mixed model analysis of individual participant data

Model	Variable	Original results						After retransformation[a]		
		Estimate	SE[b]	95% CI[c]		P-value	I^2 (%)	Estimate	95% CI[c]	
Unadjusted	ln (WTA/WTP)	0.369	0.104	0.165	0.573	<0.01	88	1.862	1.519	2.284
Adjusted	ln (WTA/WTP)[d]	0.281	0.090	0.106	0.457	<0.01	91	1.696	1.422	2.022
	Age	0.003	0.001	0.001	0.006	0.01				
	Gender (female)	0.016	0.033	-0.049	0.081	0.63				
	Income category (low)	0.131	0.054	0.026	0.237	0.01				

[a] The estimate and CI were retransformed to the original scale with a smearing factor [42]

[b] SE: standard error

[c] CI: Confidence interval

[d] This estimate is for men aged 50 in the highest income category (=reference levels of the variables)

Sensitivity analysis: zero WTP and/or WTA values

The merged dataset contained 445 subjects (14%) with a WTA, WTP or both WTA and WTP of zero (table 4). Table 7 displays the results of the first sensitivity analysis. These results have been obtained in the same way as the results in table 5, only with exclusion of the 445 subjects reporting zero WTA and/or WTP. This analysis shows that the exclusion of zero WTA and/or WTP, generally, resulted in lower WTA/WTP ratios, with this effect being most pronounced for the mean and median WTA/WTP ratios obtained from average WTA and WTP at study level compared to those obtained from median WTA and WTP at study level. Furthermore, unsurprisingly, the impact was largest in the studies with more subjects reporting zero WTP.

Table 8 displays the results of the second sensitivity analysis, the mixed model analysis with replacement of zero values with either a ½, ⅓ or ¼ of the smallest value reported in the study from which the subjects reporting zero WTA and/or WTP originated from. These results have been obtained in a similar way as the results in table 6. The estimated WTA/WTP ratios were much larger when zeroes were replaced by a small value compared to when zeroes were excluded from the analysis. This may partly be caused by the large smearing factors in the sensitivity analyses (3.7-5.7 in the sensitivity analyses versus 1.3 in the original analysis) caused by the artificial "spike" at the lower end of the distribution because of the imputation of zeroes with small values. The estimated WTA/WTP ratios were larger when the replacement values were smaller.

Table 7: WTP, WTA and WTA/WTP estimates based on individual participant data (converted to 2017 Euros), with exclusion of subjects reporting zero WTA and/or WTP

First author (year)	**N subjects reporting zero values (%)**[a]	**WTP**			**WTA**			**WTA/WTP**[b]		
		N	Mean (SD[c])	Median (quartiles)	N	Mean (SD)	Median (quartiles)	N[d]	Mean	Median
van den Berg (2005) [31]	24 (9)	219	11.17 (5.23)	10.33 (8.85|11.80)	226	12.30 (6.84)	11.80 (8.85|14.75)	246	1.10	1.14
Borisova (2003) [32]	182 (60)	121	6.41 (7.96)	3.78 (1.63|7.98)	121	8.30 (8.11)	6.30 (2.21|12.60)	121	1.30	1.67
Chiwaula (2016) [33]	1 (1)	91	19.03 (23.85)	10.25 (6.15|20.50)	91	37.53 (36.59)	24.60 (14.35|51.25)	91	1.97	2.40
Martin-Fernandez (2010) [37]	45 (10)	405	28.24 (22.94)	23.94 (10.64|37.24)	404	49.50 (29.14)	47.22 (30.59|60.52)	405	1.75	1.97
Martin-Fernandez (2013) [38]	83 (13)	570	21.06 (19.33)	12.80 (6.40|25.60)	568	27.31 (21.83)	25.60 (12.80|38.40)	570	1.30	2.00
de Meijer (2010) [39][e]										
Suppliers	62 (6)	698	12.13 (7.00)	11.44 (8.58|14.29)	794	13.68 (8.15)	11.44 (8.58|14.29)	930	1.13	1.00
Patients	47 (14)	266	10.00 (6.33)	8.58 (5.72|11.44)	260	12.07 (8.03)	11.44 (5.72|14.29)	298	1.21	1.33
Mean								2,661	1.39	1.64
Weighted average								2,661	1.37	1.64
Median								2,661	1.30	1.65

[a] These subjects were excluded from this analysis. Apart from this, the results in this table have been obtained in the same way as the results in Table 5. See last section methods for more information.

[b] Calculated from study-level WTA and WTP: mean WTA/WTP= mean WTA/mean WTP; median WTA/WTP= median WTA/median WTP

[c] SD: standard deviation

[d] Subjects with either WTA or WTP missing were still included in this analysis. Therefore, this N is higher than the N for WTP and WTA separately.

[e] As informal caregivers reported WTP and WTA for one hour extra per week and patients reported WTP and WTA for one hour extra per day, the good valued is not comparable between these groups. Therefore, the subgroups are reported separately.

Table 8: WTA/WTP ratios obtained from the mixed model analysis with replacement of zero values

	Model	**Variable**	**Original results**						**After retransformation**[a]		
			Estimate	SE[b]	95% CI[c]		P-value	I^2 (%)	Estimate	95% CI[c]	
Zero= 1/2 of the smallest value in de dataset	Unadjusted	ln (WTA/WTP)	0.711	0.223	0.273	1.149	<0.01	88	7.538	4.867	11.675
	Adjusted	ln (WTA/WTP)[d]	0.561	0.241	0.090	1.033	0.02	86	6.475	4.040	10.377
		Age	<0.001	0.003	-0.006	0.006	0.96				
		Gender (female)	0.025	0.046	-0.065	0.115	0.59				
		Income category (low)	0.269	0.071	0.129	0.408	<0.01				
Zero= 1/3 of the smallest value in de dataset	Unadjusted	ln (WTA/WTP)	0.750	0.241	0.278	1.222	<0.01	88	10.088	6.292	16.175
	Adjusted	ln (WTA/WTP)[d]	0.590	0.259	0.082	1.096	0.02	86	8.550	5.149	14.200
		Age	<-0.001	0.004	-0.007	0.006	0.95				
		Gender (female)	0.026	0.049	-0.070	0.122	0.59				
		Income category (low)	0.292	0.075	0.144	0.440	<0.01				
Zero= 1/4 of the smallest value in de dataset	Unadjusted	ln (WTA/WTP)	0.778	0.254	0.281	1.275	<0.01	88	10.371	6.310	17.048
	Adjusted	ln (WTA/WTP)[d]	0.608	0.272	0.0756	1.141	0.03	86	10.557	6.196	17.987
		Age	<-0.001	0.004	-0.008	0.007	0.95				
		Gender (female)	0.027	0.051	-0.073	0.127	0.59				
		Income category (low)	0.308	0.079	0.154	0.462	<0.01				

[a] The estimate and CI were retransformed to the original scale with a smearing factor [42]

[b] SE: standard error

[c] CI: Confidence interval

[d] This estimate is for men aged 50 in the highest income category (=reference levels of the variables)

Discussion

The aim of this study was to review the available evidence on the disparity between WTA and WTP for healthcare goods and services using a comprehensive tiered approach consisting of 1) a systematic review, 2) an aggregate data meta-analysis (AD-MA), and 3) an individual participant data meta-analysis (IPD-MA). In the AD-MA, we found an average WTA/WTP ratio of 1.73 (median 1.31) for mean estimates and of 1.58 (median 1.00) for median estimates. In the IPD-MA, we found an uncorrected WTA/WTP ratio of 1.86 (95% confidence interval: 1.52-2.28) and a WTA/WTP ratio adjusted for age, gender and income of 1.70 (95% confidence interval: 1.42-2.02). The approach to deal with zero WTP and/or WTA values considerably impacted the WTA/WTP ratio found.

This study found a significant effect of income category and age on the WTA/WTP ratio. No effect of gender was found. As previous meta-analyses on WTA and WTP have not tested the effect of age, gender and income on the WTA/WTP ratio, it is not possible to compare these findings with other studies. However, these findings seem to correspond with the well-known income effect, that says that because WTP is constrained by income while WTA is not, there may be a substantial disparity between WTA and WTP when 1) the change concerned is large, 2) the value of the good concerned is high, or 3) the income elasticity for the good concerned is high and increasing with income [5, 15]. The reason for this is that when the value of the good increases, the WTP will increase until the income constraint is reached, while WTA would become infinite. As people with lower incomes have a lower income constraint than people with higher incomes, the WTA-WTP disparity should be larger for people with lower incomes than for people with higher incomes, as was indeed was found in this study.

To get an impression of the impact of our approach to zero WTP and/or WTA responses in our main analyses, we have conducted two sensitivity analyses. The results of these sensitivity analyses indicate that the approach to dealing with subjects reporting zero WTP and/or WTA may considerably affect the WTA/WTP ratio. To our knowledge, the issue on how to deal with zero WTP and WTA has not received much attention in the scientific literature so far. To determine the best approach to dealing with zero responses, it is important to know the rationale behind reporting zeroes in stated preference studies. Qualitative inquiry during or directly after the administration of the WTP and WTA task may provide more insight in the reasons behind zero responses and subsequently give guidance on the most valid approach of dealing with zero responses (which may be another approach than was used in this meta-analysis). Some studies already included follow-up questions when eliciting WTP and found that zero responses may be protest responses as well as real zeroes [27-29]. However, more research on the rationale behind zero responses and the best approach to deal with these zero responses in the analysis is warranted. Furthermore, in order to prevent analysis and interpretation problems with regards to zero WTP and/or WTA such as encountered in our review, we recommend future research to decrease the number of zero responses by using other contingent valuation methods than open-ended questions, as previous reviews have shown that open-ended question formats are more prone to zero responses than other contingent valuation methods [2, 43]. Moreover, when using a closed-ended question format, researchers are recommended not to include the value zero in the option list, but, instead, to only provide the option ‘the good is not worth anything to me’. This will force subjects to think twice before reporting a zero, which will decrease the number of non-true zero responses. For the remaining zero responses, in order to determine how to best handle these individual zero responses in the analysis (e.g. exclusion or imputation), researchers are recommended to include a probing question which pops-up if respondents report zero WTP and/or WTA. Answer options should at least cover the following possible reasons underlying zero responses: not understanding the question, protest response, value of the good is smaller than the answer option provided, and true zero (‘the good is not worth anything to me’). Including such a probing question, will open the ‘black box’ of zero responses, facilitating the decision on how to deal with individual zero responses in the analysis, and will force subjects to think about their zero response which may, in some cases, result in subjects changing their zero into their true value.

Comparison with previous studies

The WTA/WTP ratios found in our meta-analysis are considerably lower than those found in previous meta-analyses/reviews. A possible explanation of this may be that one of the studies included in the review by O’Brien et al., was not included in our meta-analysis as it was not identified in our search because title and abstract did not contain WTA or variations thereof. This study reported a very high WTA/WTP ratio of 6.4 for a non-fatal injury which may be explained by the fact that the change valued in the WTA scenario (i.e. no injury vs full injury) was larger than the change valued in the WTP scenario (i.e. small injury vs full injury) [14]. Hence, it may not be surprising that this ratio is much larger than the ratios found in our meta-analysis. The estimate for health and safety goods in the meta-analysis by Tunçel and Hammitt was obtained from 11 studies of which 7 were not included in our meta-analysis. These seven studies reported generally larger WTA/WTP ratios than the studies included in our meta-analysis and predominantly valued traffic safety, job safety and product safety, i.e. safety goods [10]. This indicates that the WTA/WTP disparity may be larger in safety studies than in health studies, which both may explain why our meta-analysis found a smaller disparity and stresses the need for a separate WTA/WTP estimate for healthcare goods and services, as has been obtained in our meta-analysis.

Another possible explanation for the relatively small WTA-WTP disparity found in our review may be that the studies included in our review valued relatively small changes in healthcare goods and services, such as one hour of informal care or one general practitioner consultation. According to standard economic theory, due to declining marginal utility, the WTA/WTP ratio is an increasing function of the size of the change valued [44]. As a consequence, the WTA/WTP ratio is anticipated to be larger when the changes in healthcare goods and services to be valued are truly substantive, such as a year of informal care or an orphan drug. In order to assess the degree to which the WTA/WTP ratio for healthcare goods and services is an increasing function of the size of the change valued, we recommend future research to estimate the WTA/WTP ratio for differently sized changes in the healthcare good or service concerned.

Furthermore, another possible explanation for the relatively low WTA/WTP ratio found in our review may be that subjects were quite familiar with the goods being valued. Three studies asked informal caregivers and/or informal care recipients to value informal care. Furthermore, two studies valued primary care (GP or nurse), which is a type of care many people are familiar with. If people are more familiar with the goods they value, they are more certain about their preferences and therefore report WTA and WTP values that are closer together [45]. Besides, many studies in this meta-analysis elicited WTA and WTP in the same questionnaire. Therefore, subjects could have used one of the measures as a reference for the other.

Implications of our findings

The results of this study imply that losses in healthcare goods or services are valued somewhat differently from similarly sized gains in healthcare goods and services. This may have implications for cost-benefit analyses of healthcare interventions. In cost-benefit analyses, the welfare effect of healthcare interventions is transformed into monetary units using the WTP for gains in healthcare and the WTA for losses in healthcare. However, as shown, losses in healthcare get a different weight than gains. There has been a lot of debate across different economic sectors on whether WTA or WTP should be used in the context of losses. Some authors, such as those from the NOAA panel on contingent valuation, argue that WTP should always be used because WTA is biased and WTP constitutes a more conservative estimate of welfare change [46]. Others argue that WTA is valid and, hence, that the most accurate measure of welfare change depends on the direction of the change from the reference point [47, 48]. This debate is still ongoing, and our study does not provide any conclusive answers to resolve this issue.

Furthermore, our findings may have implications for reimbursement decision-making based on cost-effectiveness/cost-utility analyses. Although the effects of healthcare goods and services are expressed in health units in cost-effectiveness analyses and in Quality Adjusted Life Years (QALYs) in cost-utility analyses, WTA and WTP still need to be used in order to make reimbursement decisions based on these analyses. In many countries, implicit or explicit thresholds for the WTP for additional health outcomes have been used in reimbursement decision-making. For instance, NICE in England and Wales uses a threshold of £20,000 to £30,000 per QALY gained [49, 50], and the National Health Care Institute in the Netherlands uses a threshold of €20,000 to €80,000 per QALY gained, depending on disease severity [51]. However, a threshold for the WTA for a loss in health does not exist. Therefore, the WTP threshold has often been used for such decisions [52]. However, as our study shows, the WTA for healthcare goods and services is somewhat higher than WTP. Therefore, to align policy with societal preferences, one might argue to use a somewhat higher threshold in the domain of losses compared to the domain of gains. To this end, Severens et al. (2005) suggested to use a modified cost-effectiveness acceptability curve approach to provide insight in the impact of the WTA-WTP disparity on the probability of an intervention being cost-effective. This information could then be incorporated in reimbursement decision-making, facilitating a societal debate on this issue [53]. However, others have suggested that the same threshold should be used for decisions in the context of gains and losses, as using different thresholds may introduce substantial inefficiencies in the allocation of the healthcare budget [54-56]. Hence, whether the WTA-WTP disparity should be incorporated in healthcare policymaking is a political trade-off between aligning policy with societal preferences on the one hand, and stimulating efficiency in the allocation of healthcare budgets on the other hand.

Besides, the results of this study can also be used to better understand problems with disinvestment, which is the full/partial withdrawal of the reimbursement of healthcare interventions [57]. Decisions on disinvestment have often been perceived to be much more difficult than decisions on (not) starting reimbursement of healthcare [58, 59], a phenomenon that has also been observed in the context of conditional reimbursement [21]. In this study, we found a small disparity between WTA and WTP, implying that, in the healthcare context, people attach more value to losses than to gains. This may also partly explain the perceived difficulty of disinvestment compared to investment as the former is in the domain of losses and the latter is in the domain of gains.

Strengths and limitations

In this study, we used a systematic approach to estimate the WTA/WTP ratio for healthcare goods and services. The eligibility criteria were strictly applied to derive WTA and WTP estimates that were based on a similar change and elicited in the same way. In this way, we ensured that the WTA/WTP ratios derived were not biased by incomparable WTA and WTP scenarios. Furthermore, by combining data from different studies in our meta-analysis, we were able to obtain a higher level of evidence and more insight in the uncertainty surrounding the disparity between WTA and WTP than previous studies did.

Our study, however, also has some limitations. First, the studies included in our meta-analysis were quite heterogeneous as different (changes in) healthcare goods and services were valued by different subject groups using different elicitation and administration methods. Furthermore, studies were conducted in different settings. Due to the small number of studies available, we were not able to test the effect of these different settings and methods on the WTA/WTP ratio for healthcare goods and services. Therefore, more studies on the WTA/WTP ratio for healthcare goods and services are needed in order to obtain more insight in this issue.

Secondly, as we have not tested the quality of the included studies, we were not able to weight the study estimates based on their quality. However, we are not aware of any quality assessment instrument applicable to WTA/WTP studies, hampering the incorporation of study quality in the analyses.

Thirdly, although we were able to include the largest studies from our review in the IPD-MA, the number of studies included in this meta-analysis is still quite small. Furthermore, most studies included in the IPD-MA valued informal care or primary care services. Therefore, our results cannot be generalized to all healthcare goods and services in general. More research is needed to obtain insight in the WTA/WTP ratio for a broader range of healthcare goods and services.

Fourthly, in the mixed model we calculated the WTA/WTP ratio using the mean of ratios approach. We are aware that using the ratio of means approach instead, could have resulted in a different estimate of the WTA/WTP ratio [60, 61]. However, due to differences in the goods and services valued in the included studies, we were not able to use the ratio of means approach and to determine the effect of using one approach over the other on the WTA/WTP ratio.

Fifthly, in our analysis we assumed the association between age and ln(WTA/WTP) to be linear. However, some studies showed small deviations from this assumption. Nonetheless, as correcting for non-linearity would not result in significantly improved model fits, we decided not to correct for this applying the credo: “as simple as possible, as complex as necessary”.

Finally, we have used the median as cut-off point to transform the income data to two categories. Although, there was no better option to combine the income data, this approach may have hampered the interpretation of the effect of income. The reason for this is that the study population may not reflect the general population in terms of income. For instance, in the study on the valuation of methadone maintenance, it is imaginable that the respondents had a relatively low income. The implication of this would be that our income categories based on median income actually represent very low versus quite low income.

Conclusions and recommendations

This study found aggregated WTA/WTP ratios between 1.58 and 1.86 for healthcare goods and services, indicating that losses are weighted somewhat differently from gains. The ratio found depends on the method used to calculate the WTA/WTP ratio and the approach on how to deal with subjects reporting zero WTP and/or WTA. Irrespective of the method used, the ratios found in our meta-analysis were smaller than the ratios found in previous meta-analyses. For this reason, the WTA-WTP disparity in the healthcare sector may be less of a problem than what was thought based on previous studies. However, we cannot exclude the possibility that the relatively small disparity found, is related to the fact that the studies in our review valued relatively small gains and losses in healthcare goods and services, with which subjects were quite familiar. Future empirical work may explicitly test the effect of size of the change valued on the WTA/WTP ratio through a within-person assessment of differently sized changes in healthcare goods and services. Furthermore, we recommend future research to pay attention to the reasons behind zero WTA and WTP responses and the best methodological way of dealing with these responses in the analysis.

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Appendices

Appendix A: Search strategies

Embase and Medline

#1	wta:ti,ab,de OR ‘willingness to accept’:ti,ab,de OR (health NEAR/5 forgone):ti,ab,de OR (health NEAR/5 foregone):ti,ab,de OR (qaly* NEAR/5 forgone):ti,ab,de OR (qaly* NEAR/5 foregone):ti,ab,de OR (‘quality adjusted life’ NEAR/5 forgone):ti,ab,de
#2	wtp:ti,ab,de OR ‘willingness to pay’:ti,ab,de OR ‘willingness-to-pay’:ti,ab,de
#3	#1 AND #2
#4	#3 AND (english:la OR dutch:la)

Scopus

#1	TITLE-ABS-KEY(WTA OR (willingness-to-accept) OR (health W/5 forgone) OR (health W/5 foregone) OR (qaly* W/5 forgone) OR (qaly* W/5 foregone) OR (quality-adjusted-life W/5 forgone)) OR TITLE-ABS-KEY((willingness W/5 accept))
#2	TITLE-ABS-KEY(WTP OR (willingness-to-pay))
#3	#1 AND #2
#4	TITLE-ABS-KEY(health OR healthcare OR care* OR welfare OR disease* OR practitioner* OR specialist* OR (family-physician*) OR medication* OR hospital* OR patient* OR clinical* OR treatment OR therapy OR chemotherapy OR pharmaceut* OR screening OR testing OR vaccin* OR prevention OR preventive)
#5	#3 AND #4
#6	LANGUAGE(english OR dutch)
#7	#5 AND #6

SciSearch

S1	AB,TI(WTA OR (willingness-to-accept) OR (health NEAR/5 forgone) OR (health NEAR/5 foregone) OR (qaly* NEAR/5 forgone) OR (qaly* NEAR/5 foregone) OR (“quality-adjusted-life” NEAR/5 forgone) OR (willingness NEAR/5 accept)) OR IF(WTA OR (willingness-to-accept) OR (health NEAR/5 forgone) OR (health NEAR/5 foregone) OR (qaly* NEAR/5 forgone) OR (qaly* NEAR/5 foregone) OR (“quality-adjusted-life” NEAR/5 forgone) OR (willingness NEAR/5 accept))
S2	AB,TI(WTP OR (willingness-to-pay)) OR IF(WTP OR (willingness-to-pay))
S3	S1 AND S2
S4	AB,TI(health OR healthcare OR care* OR welfare OR disease* OR practitioner* OR specialist* OR (family-physician*) OR medication* OR hospital* OR patient* OR clinical* OR treatment OR therapy OR chemotherapy OR pharmaceut* OR screening OR testing OR vaccin* OR prevention OR preventive) OR IF (health OR healthcare OR care* OR welfare OR disease* OR practitioner* OR specialist* OR (family-physician*) OR medication* OR hospital* OR patient* OR clinical* OR treatment OR therapy OR chemotherapy OR pharmaceut* OR screening OR testing OR vaccin* OR prevention OR preventive)
S5	S3 AND S4
S6	la(english or dutch)
S7	S5 AND S6

Econlit

1	(WTA or “willingness to accept” or willingness-to-accept or (health adj5 forgone) or (health adj5 foregone) or (qaly* adj5 forgone) or (qaly* adj5 foregone) or (quality adjusted life adj5 forgone)).tw.
2	(WTA or “willingness to accept” or willingness-to-accept or (health adj5 forgone) or (health adj5 foregone) or (qaly* adj5 forgone) or (qaly* adj5 foregone) or (quality adjusted life adj5 forgone)).kw.
3	1 or 2
4	(WTP or “willingness to pay” or willingness-to-pay).tw. or (WTP or “willingness to pay” or willingness-to-pay).kw.
5	3 and 4
6	(health OR healthcare OR care* OR welfare OR disease* OR practitioner* OR specialist* OR (family-physician*) OR medication* OR hospital* OR patient* OR clinical* OR treatment OR therapy OR chemotherapy OR pharmaceut* OR screening OR testing OR vaccin* OR prevention OR preventive).tw. OR (health OR healthcare OR care* OR welfare OR disease* OR practitioner* OR specialist* OR (family-physician*) OR medication* OR hospital* OR patient* OR clinical* OR treatment OR therapy OR chemotherapy OR pharmaceut* OR screening OR testing OR vaccin* OR prevention OR preventive).hw.
7	5 and 6
8	7 and (english or dutch).lg.

Appendix B: Study characteristics extracted in the systematic review

Extracted study characteristics	Categories (if applicable)	Comment
First author		
Year		Year of publication
Country		
Good/service valued		
Number of subjects		
Subject sample type	General public (including subjects at which good is aimed)	
	General public (excluding subjects at which good is aimed)	
	Patients/clients	
	Parents/guardians of patients/clients	
	Suppliers	
Within/between subject	Within	Whether it concerns a within subject design (subjects answer both the WTA and WTP question) or a between subject design (subjects are randomized to either the WTA or WTP question)
	Between	
Elicitation method	Payment card	
	Discrete choice experiment	
	Open-ended single question	
	Close-ended single question	
	Close-ended question with open-ended follow-up	
	Iterated close-ended questions (bidding game)	
Administration method	Interviews	If possible, make a distinction between telephone interviews and (face-to-face) interviews
	Surveys	If possible, make a distinction between supervised and not supervised surveys
Payment vehicle	Out-of-pocket	
	Tax	
	Insurance premium	
	Public	A payment from/to the government budget
Payment frequency	Hourly	
	Daily	
	Monthly	
	Annually	
	Once	
	Per visit	

Appendix C: Studies included in the review and meta-analysis

Study reference	Included in review	Included in AD-MA	Included in IPD-MA
Bayen, E., Jourdan, C., Ghout, I., Darnoux, E., Azerad, S., Vallat-Azouvi, C., et al. (**2016**). Objective and subjective burden of informal caregivers 4 years after a severe traumatic brain injury: Results from the Paris-TBI study. Journal of Head Trauma Rehabilitation, 31(5), E59-E67. doi:10.1097/HTR.0000000000000079	X	X	
van den Berg, B., Bleichrodt, H., & Eeckhoudt, L. (**2005**). The economic value of informal care: A study of informal caregivers' and patients' willingness to pay and willingness to accept for informal care. Health Economics, 14(4), 363-376. doi:10.1002/hec.980	X	X	X
Borisova, N. N., & Goodman, A. C. (**2003**). Measuring the value of time for methadone maintenance clients: Willingness to pay, willingness to accept, and the wage rate. Health Economics, *12*(4), 323-334. doi:10.1002/hec.738	X	X	X
Chiwaula, L. S., Chirwa, G. C., Caltado, F., Kapito-Tembo, A., Hosseinipour, M. C., van Lettow, et al. (**2016**). The value of informal care in the context of option B+ in Malawi: a contingent valuation approach. BMC Health Services Research, 16, 136. doi:10.1186/s12913-016-1381-y	X	X	X
Finkelstein, E., Malhotra, C., Chay, J., Ozdemir, S., Chopra, A., & Kanesvaran, R. (**2016**). Impact of Treatment Subsidies and Cash Payouts on Treatment Choices at the End of Life. Value in Health, 19(6), 788-794. doi:10.1016/j.jval.2016.02.015	X	X	
Grutters, J. P. C., Kessels, A. G. H., Dirksen, C. D., Van Helvoort-Postulart, D., Anteunis, L. J. C., & Joore, M. A. (**2008**). Willingness to accept versus willingness to pay in a discrete choice experiment. Value in Health, 11(7), 1110-1119. doi:10.1111/j.1524-4733.2008.00340.x	X	X	
Manan, M. M., Ali, S. M., Khan, M. A. N., & Jafarian, S. (**2015**). Estimation of out-of-pocket costs of patients at the Methadone Maintenance therapy clinic in Malaysia. *Pakistan Journal of Pharmaceutical Sciences, 28*(5), 1705-1711.	X	X	
Martín-Fernández, J., del Cura-González, M. I., Gómez-Gascón, T., Oliva-Moreno, J., Domínguez-Bidagor, J., Beamud-Lagos, M., & Pérez-Rivas, F. J. (**2010**). Differences between willingness to pay and willingness to accept for visits by a family physician: a contingent valuation study. *BMC Public Health, 10*, 236. doi:10.1186/1471-2458-10-236	X	X	X
Martín-Fernández, J., del Cura-González, M. I., Rodríguez-Martínez, G., Ariza-Cardiel, G., Zamora, J., Gómez-Gascón, et al. (**2013**). Economic Valuation of Health Care Services in Public Health Systems: A Study about Willingness to Pay (WTP) for Nursing Consultations. PloS One, 8(4), e62840. doi:10.1371/journal.pone.0062840	X	X	X
de Meijer, C., Brouwer, W., Koopmanschap, M., Van Den Berg, B., & Van Exel, J. (**2010**). The value of informal care - A further investigation of the feasibility of contingent valuation in informal caregivers. Health Economics, 19(7), 755-771. doi:10.1002/hec.1513	X	X	X

Study reference	**Included in review**	**Included in AD-MA**	**Included in IPD-MA**
O'Brien, B. J., Goeree, R., Gafni, A., Torrance, G. W., Pauly, M. V., Erder, H., et al. (**1998**). Assessing the value of a new pharmaceutical. A feasibility study of contingent valuation in managed care. Medical Care, 36(3), 370-384.	X	X	
Tsuji, M., & Suzuki, W. (**2004**). The application of CVM for assessing the tele-health system: an analysis of the discrepancy between WTP and WTA based on survey data. In C. D. Aliprantis, K. J. Arrow, P. Hammond, F. Kubler, H.-M. Wu, & N. C. Yannelis (Eds.), Assets, Beliefs, and Equilibria in Economic Dynamics: Essays in Honor of Mordecai Kurz (pp. 493-506). Berlin, Heidelberg: Springer Berlin Heidelberg.	X	X	
Whynes, D. K., & Sach, T. H. (**2007**). WTP and WTA: Do people think differently? Social Science and Medicine, 65(5), 946-957. doi:10.1016/j.socscimed.2007.04.014	X	X	

Appendix D: WTP, WTA and WTA/WTP estimates based on original individual participant data (not converted)

First author (year of publication)	Currency (year of data collection)	WTP			WTA			WTA/WTP[a]		
		N	Mean (SD)	Median (quartiles)	N	Mean (SD)	Median (quartiles)	N[b]	Mean	Median
van den Berg (2005)	Dutch guilders (2001)	241	17.46 (9.91)	15.00 (10.00|20.00)	246	20.05 (12.19)	17.75 (15.00|25.00)	270	1.15	1.18
Borisova (2003)	US Dollars (1999)	303	2.47 (4.95)	0.00 (0.00|3.00)	303	7.73 (10.79)	65.00 (0.88|10.00)	303	3.13	NA[c]
Chiwaula (2016)[d]	Malawian Kwatcha (2013)	92	9.18 (11.60)	5.00 (3.00|10.00)	92	18.10 (17.85)	12.00 (7.00|23.75)	92	1.97	2.40
Martin-Fernandez (2010)	Euros (2008)	451	19.62 (17.47)	18.00 (8.00|28.00)	450	36.04 (22.58)	35.50 (18.00|45.50)	451	1.84	1.97
Martin-Fernandez (2013)	Euros (2013)	653	14.38 (15.12)	10.00 (5.00|20.00)	648	20.90 (17.13)	20.00 (10.00|30.00)	653	1.45	2.00
de Meijer (2010)[e]										
Suppliers	Euros (2002)	753	9.12 (5.80)	9.08 (6.81|11.34)	849	10.48 (6.75)	9.08 (6.81|11.34)	992	1.15	1.00
Patients	Euros (2002)	311	6.90 (5.34)	6.81 (4.54|9.08)	300	8.99 (6.49)	8.17 (4.54|11.34)	345	1.30	2.30
Mean								3,106	1.71	1.63
Weighted average								3,106	1.70	1.63
Median								3,106	1.45	1.59

[a] Calculated from study-level WTA and WTP: mean WTA/WTP= mean WTA/mean WTP; median WTA/WTP= median WTA/median WTP

[b] Subjects with either WTA or WTP missing were still included in this analysis. Therefore, this N is higher than the N for WTP and WTA separately.

[c] Median WTP is zero. Therefore, the WTA/WTP ratio based on medians could not be calculated for this study.

[d] WTP and WTA values are reported in thousand Malawian Kwatcha

[e] As informal caregivers reported WTP and WTA for one hour extra per week and patients reported WTP and WTA for one hour extra per day, the good valued is not comparable between these groups. Therefore, the subgroups are reported separately.

Appendix E: Univariate results

Subject group		N	WTA/WTP ratio				
			Mean	**SD**	**Median**	**Quartiles**	
Income category	Low	902[a]	2.24	3.15	1.00	1.00	2.00
	High	932[a]	1.57	1.91	1.00	1.00	1.40
Gender	Female	1295	1.88	2.46	1.00	1.00	1.97
	Male	837	1.87	2.62	1.00	1.00	1.97
Age category[b]	<45 years	485	1.94	2.59	1.00	1.00	2.00
	45-56 years	531	1.60	1.53	1.00	1.00	1.67
	57-70 years	559	1.77	2.47	1.00	1.00	1.67
	>70 years	598	2.13	3.08	1.00	1.00	2.00

[a] Not all studies provided income data. Therefore, the number of subjects in these categories do not add up to 2173

[b] Categories are based on the quartiles of age in de overall dataset

Appendix F: WTA-WTP for different groups of subjects

Table F1: multivariate WTA/WTP estimates for different levels of subject characteristics

Age	Subject group	ln(WTA/WTP)	WTA/WTP	95% confidence interval	
30	Income= low	0.352	1.82	1.46	2.26
30	Income= high	0.221	1.60	1.34	1.90
30	Female	0.294	1.72	1.40	2.11
30	Male	0.279	1.69	1.42	2.02
65	Income= low	0.472	2.05	1.58	2.66
65	Income= high	0.341	1.80	1.47	2.20
65	Female	0.415	1.94	1.52	2.47
65	Male	0.399	1.91	1.54	2.35

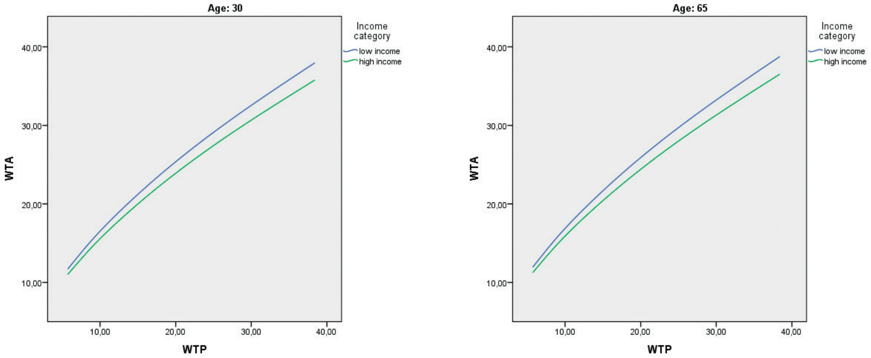


Figure F1: WTA and WTP for people with low and high income, aged 30 and 65

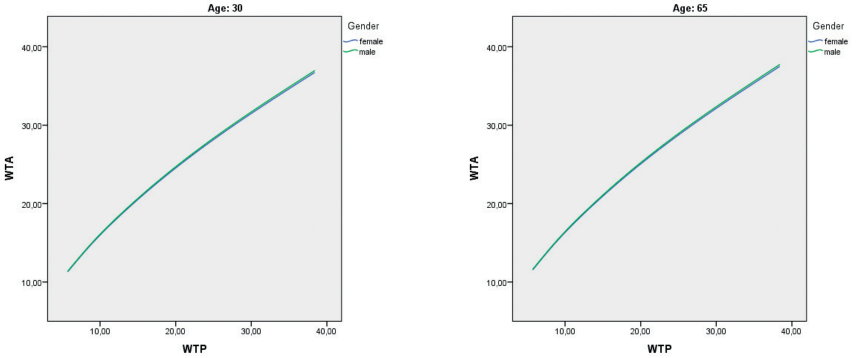


Figure F2: WTA and WTP for males and females, aged 30 and 65

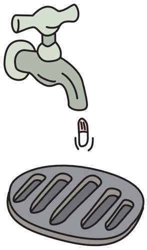
6

If You Were A Policymaker, Which Treatment Would You Disinvest?

A participatory value evaluation on public preferences for active disinvestment of healthcare interventions in the Netherlands

Rotteveel AH
Lambooij MS
Over EAB
Hernández JI
Suijkerbuijk AWM
De Blaeij AT
de Wit GA
Mouter N

This chapter has been submitted as: Rotteveel AH, Lambooij MS, Over EAB, Hernandez JI, Suijkerbuijk AWM, de Blaeij AT, et al. If you were a policymaker, which treatment would you disinvest? A participatory value evaluation on public preferences for active disinvestment of healthcare interventions in the Netherlands



Abstract

Introduction: Currently, it is not known what attributes of healthcare interventions citizens consider important in disinvestment decision-making (i.e. decisions to discontinue reimbursement). Therefore, this study aims to investigate the preferences of citizens of the Netherlands towards the relative importance of attributes of healthcare interventions in the context of disinvestment.

Methods: A participatory value evaluation (PVE) was conducted in April and May 2020. In this PVE, 1143 Dutch citizens were asked to save at least €100 million by selecting healthcare interventions for disinvestment from a list of eight unlabelled healthcare interventions, described solely with attributes. A portfolio choice model was used to analyse participants' choices.

Results: Participants preferred to disinvest healthcare interventions resulting in smaller gains in quality of life and life expectancy that are provided to older patient groups. Portfolio's (i.e. combinations of healthcare interventions) resulting in smaller savings were preferred for disinvestment over portfolio's with larger savings.

Conclusion: The disinvestment of healthcare interventions resulting in smaller health gains and that are targeted at older patient groups are likely to receive most public support. By incorporating this information in the selection of candidate interventions for disinvestment and the communication on disinvestment decisions, policymakers may increase public support for disinvestment.

Introduction

In many countries, the large increase in healthcare expenditures [1] has put a considerable pressure on public financing [2, 3]. To limit the increase in healthcare expenditures, policymakers have increasingly and more strictly been assessing the reimbursement of new healthcare interventions, defined in this paper as a broad range of curative care, such as medicines, therapies, and medical devices.

Although the criteria for the reimbursement of new healthcare interventions differ between countries and settings [4, 5], previous studies have shown that clinical effectiveness (i.e. health gain of the intervention) is an important criterion in many countries [5, 6]. Nevertheless, it has been estimated that 20 to 30% of healthcare costs is spent on interventions that are not effective [7, 8]. This may, for instance, be due to the less strict application of effectiveness as a criterion in past reimbursement decisions or to new evidence indicating that healthcare interventions may be less effective than thought before [9, 10]. The large amount of ineffective healthcare interventions currently being used and reimbursed suggests that a lot can be gained both in terms of health and healthcare expenditures from the structured, evidence-based reassessment of these interventions, which has been called Health Technology Reassessment (HTR) [11, 12]. HTR, just as reimbursement decision-making, has often been based on more considerations than effectiveness alone, such as the economic, social and ethical aspects of an intervention [5, 12]. If, as a result of HTR, it is concluded that a healthcare intervention does not meet the applicable criteria, it may be decided to withdraw the reimbursement of this intervention. The withdrawal of reimbursement is called disinvestment [9, 13].

Despite the potential merit of HTR with regard to health gains and containing healthcare expenditures, the actual reassessment of healthcare interventions is rare. Over a period of ten years, 34 healthcare interventions were reassessed in the Netherlands [14]. This is just a very small fraction of the healthcare interventions that are currently provided to patients. A possible explanation for the lack of HTR’s, can be found in the difficulty to disinvest healthcare interventions [12, 15]. Support from healthcare providers, policymakers and the general public has been shown to be essential for successful disinvestment [13, 14]. In this study, we focus on public support. Insight in which attributes (i.e. characteristics) of healthcare interventions citizens find most important for policymakers to take into account in disinvestment decisions may help policymakers to propose disinvestment decisions that receive more public support.

Much evidence is available on the relative importance of attributes in the context of the assessment of new healthcare interventions for reimbursement (investment decisions). A systematic review concluded that studies consistently found that health gain was considered most important by citizens. Both gain in quality of life and gain in life expectancy have been shown to be important, with citizens making trade-offs between them [16]. The importance attached to health gains may be different, depending on the health state before and after treatment [16, 17]. In addition to health gain, age of the patient group has also shown to be important [16]. Previous research on disinvestment policy processes indicated that different considerations may be relevant in the context of disinvestment compared to the context of investment [10, 14]. Therefore, it is not clear whether these attributes are also considered important in the context of disinvestment.

Evidence on attributes that citizens find relevant in the context of disinvestment is limited to four studies [18-22]. These studies found that citizens consider it important to take medical necessity, effectiveness, real-world cost(-effectiveness) data, availability of treatment alternatives, disease severity, and externalities into account in disinvestment decisions [18, 20, 22]. Moreover, while one study found that citizens are reluctant to disrupt the current treatment of patients [19], no indication for this was found in another study [22]. Although these studies already provide some insight in the attributes that citizens find relevant in the context of disinvestment, it is not known what the relative importance of these attributes is.

The aim of this study is to investigate the preferences of citizens of the Netherlands towards the relative importance of attributes of healthcare interventions in the context of disinvestment. To this end, we asked a representative sample of citizens to make disinvestment decisions by selecting two or more healthcare interventions from a list of eight candidate interventions for disinvestment using the participatory value evaluation (PVE) approach. The PVE approach was used as it allows participants to make a decision as if they were a policymaker (i.e. it mimics the decision-making context), while it provides insight in the preferences citizens have with regards to the attributes of healthcare interventions as well as the allocation of public money [23] in the context of disinvestment. As previous research in the investment context showed that the importance attached to gains in health is affected by the health state before and after treatment [16], the secondary aim of this study is to assess what the effect of the health state before and after treatment is on the importance attached by Dutch citizens to a gain in health in the disinvestment context.

Methods

Here, we have used participatory value evaluation (PVE), a novel stated preference method in which participants are requested to choose the best portfolio (i.e. combination) of policy options within a given budget [24]. The policy options, in this case, the candidate interventions for disinvestment, are specified in terms of attributes and levels. From the portfolios that participants choose, preferences with regards to the attributes and the amount of public money spent/saved can be obtained [24].

Design

In an online survey, participants were presented with 1) an introduction to the PVE task, 2) the PVE task itself, and 3) questions on the sociodemographic characteristics of participants and how participants experienced the PVE task. The questions on the sociodemographic characteristics of participants were included to check the representativeness of the sample. The questions on how participants experienced the PVE task were included to learn more on the feasibility and usefulness of PVE.

In the introduction to the PVE task, participants were familiarized with the decision-problem and the task, and received instructions on the web-based environment (see Appendix A). Participants were told that, because healthcare expenditures are rising, the government asks citizens for their advice on savings in healthcare. They were asked which treatments they would disinvest in order to save at least €100 million from the healthcare budget to be able to reimburse new/other treatments. The threshold for the minimum required savings was set at €100 million, to ensure that the savings were substantial enough to make an impact (±1% of Dutch healthcare budget), while still being feasible through the disinvestment of a limited number of healthcare interventions. In the PVE task (see Appendix B), participants were shown a list of 8 hypothetical treatments that could be disinvested to achieve the €100 million of savings. These treatments were not labelled, but randomly numbered and described with attributes and levels.

Attributes and levels

To assess whether similar attributes are relevant in the investment and disinvestment context, while keeping the PVE task manageable for participants, we selected the most important attributes as identified in the investment context as attributes in the current study: gain in quality of life, gain in remaining life expectancy and age of the patient group [16, 17]. In addition, an attribute that may be specifically important in the disinvestment context was also included: availability of an alternative treatment [20, 22]. Finally, budget-impact was included as this is a required attribute for PVE. Participants were explained that gains in quality of life and remaining life expectancy as well as the budget-impact were respective to the alternative treatment, if available. To assess whether the value attached to gains in health (i.e. quality of life and life expectancy) is affected by the health state (i.e. quality of life and life expectancy) before and after treatment, these attributes were also included in the PVE. A description of how the attribute levels were determined is included in Appendix C. All attributes and levels are displayed in Table 1.

Table 1: Attributes and attribute levels

Attributes	Attribute levels
Budget-impact per year (Euros): number of patients treated * costs per patient per year[a]	1,000,000 (1,000 patients*1,000 Euros)
	1,000,000 (200 patients*5000 Euros)
	1,500,000 (15,000 patients*100 Euros)
	2,000,000 (400 patients*5,000 Euros)
	3,000,000 (15,000 patients*200 Euros)
	5,000,000 (1,000 patients*5000 Euros)
	9,000,000 (90,000 patients*100 Euros)
	10,000,000 (200 patients*50,000 Euros)
	15,000,000 (15,000 patients*1000 Euros)
	18,000,000 (90,000 patients*200 Euros)
	18,000,000 (200 patients*90,000 Euros)
	20,000,000 (400 patients*50,000 Euros)
	36,000,000 (400 patients*90,000 Euros)
	40,000,000 (400,000 patients*100 Euros)
	50,000,000 (1,000 patients*50,000 Euros)
	75,000,000 (15,000 patients*5,000 Euros)
	80,000,000 (400,000 patients*200 Euros)
	90,000,000 (90,000 patients*1,000 Euros)
	90,000,000 (1,000 patients*90,000 Euros)
Quality of life before treatment[b] (score between 0% (=death) and 100% (=full health))	35%
	55%
	75%
Quality of life after treatment[b] (score between 0% (=death) and 100% (=full health))	40%
	55%
	80%
	85%
Remaining life expectancy before treatment[c] (years)	1
	5
	15
	25
Remaining life expectancy after treatment[c] (years)	1,5
	6
	15
	30

Table 1 continued:

Attributes	**Attribute levels**
Availability of alternative treatment	An alternative treatment is available that treats the disease An alternative treatment is available that treats disease symptoms but not the disease itself This is the only available treatment for the disease and disease symptoms
Average age of the patient group involved (years)	10 35 55 75[d]

[a] The budget-impact, as well as the number of patients concerned and the costs per patient were presented to participants

[b] Quality of life before and after treatment are both displayed as separate attributes as well as combined in the attribute 'improvement in quality of life'. Quality of life before treatment ≤ Quality of life after treatment with the difference between the two being <50% (an improvement from 35% to 85% was considered unrealistic in general and if available, to not be a serious candidate for disinvestment).

[c] Life expectancy before and after treatment are both displayed as separate attributes as well as combined in the attribute 'improvement in life expectancy'. Life expectancy before treatment ≤ Life expectancy after treatment.

[d] This attribute level could only be combined with a remaining life expectancy (before and after treatment) ≤ 15 years

Experimental design

To increase the information that can be obtained from the choice models, participants must face different combinations of attribute levels for each of the eight treatments. However, collecting data for all possible combinations of attribute levels (350,208 possibilities) is unfeasible. Therefore, an experimental design containing 60 different versions was selected through a three-stage process. First, the feasible set of combinations of attribute levels was determined. Second, a candidate initial design was constructed by randomly selecting 60 combinations of feasible attribute levels, within the restrictions as defined in Table 1. Third, attribute levels of the design were iteratively changed to reduce the correlation between attributes. Through this process, combinations of attribute levels were selected that minimize the correlation between attributes, increasing the efficiency of the model estimates. A more detailed description of the evaluation metrics and algorithms used to construct the experimental design can be found in Appendix 1 of Mouter et al. 2021 [25].

Pilot testing

The PVE was pilot tested in 5 face-to-face sessions with 20 participants in total. Participants were recruited by a commercial panel organization. Groups that participate less often in panels were oversampled to assess whether the PVE method was also feasible for these participant groups. Based on the pilot tests, study materials were improved to increase the feasibility of the PVE. For instance, participants were provided with the possibility to have the text read to them (instead of reading the text themselves) and more instructions were added to the instruction video.

Data collection

Data were collected among a representative sample of the adult Dutch population with regard to age (>18 years), gender and educational level. Participants were recruited by a commercial panel organization. Prior to entering the questionnaire, they received an information letter containing information on the study purpose, questionnaire content and length, data storage, the investigator and who had access to their data, and written informed consent was obtained. Participants received a standard participation fee, following the incentive system of the panel organization. Ethical approval was obtained from the internal review board of the Erasmus School of Health Policy & Management (IRB 20-09). The research team did not have access to participants' contact information and handled the remaining participant information confidentially.

Data analysis

The econometric approach is based on the portfolio choice model [23] to estimate the preferences for attributes in PVE, and adapted for this study. Under this framework, it is assumed that participants choose to disinvest the portfolio (i.e. combination) of treatments that is least desirable to them from all other possible portfolios of treatments, given the minimum imposed savings of €100 million. The desirability of the different portfolio's depends on the perceived societal value of the treatments that are not disinvested, as well as the societal value attached to the monetary savings in addition to the imposed minimum savings of €100 million.

Let a participant n face J treatments to disinvest in the PVE. In turn, each treatment is composed by a set of K attributes and generates savings equal to the budget-impact, denoted by C_{nj}. If a treatment is chosen by the participant for disinvestment, utility decreases since the treatment is no longer available to society. However, if the participant disinvests a set of treatments that generates savings above the minimum goal explicitly imposed in the PVE – in this case 'saving at least €100 million' – denoted by B, we assume that the utility lost by disinvesting the treatments is compensated by the utility gained from the additional savings of disinvestment. Hence, the utility a participant derives from the portfolio p of non-disinvested treatments and additional savings is given by:

$$U_{np} = V_{np} + \varepsilon_{np} = \sum_{j=1}^{J} y_{nj} \cdot U_{nj} + \alpha \left(\sum_{j=1}^{J} (1 - y_{nj}) \cdot c_{nj} - B \right) + \varepsilon_{np}$$

Where Y_{nj} is a binary variable that is equal to one if the treatment is not disinvested and zero otherwise, U_{nj} is the utility derived from treatment j, α is an estimable parameter that accounts for the preferences for additional savings, and ε_{np} is an extreme-value error term. The utility of each treatment j is a function of their K attributes:

$$U_{nj} = \sum_{k}^{K} \beta_k \cdot x_{njk}$$

Where x_{njk} is the attribute level k of treatment j, and β_k is an estimable parameter that accounts for the preferences for attributes.

The choice probability of keeping the portfolio p takes the form of a multinomial logit function:

$$P_{np} = P(U_{np} \geq U_{nq}, \forall p \neq q) = \frac{e^{V_{np}}}{\sum_{q \in Q} e^{V_{nq}}}$$

Where Q is the set of all feasible portfolios of treatments. A maximum likelihood approach is used to estimate α and β, in a similar way as a standard multinomial logit model [26].

To assess whether quality of life and life expectancy before and after treatment affected the value attached to a gain in quality of life and life expectancy, two additional analyses were conducted: 1) an analysis containing both quality of life and life expectancy **before** treatment, and gain in quality of life and life expectancy as attributes, and 2) an analysis containing both quality of life and life expectancy **after** treatment, and gain in quality of life and life expectancy as attributes. As the gain in quality of life/life expectancy and the quality of life/life expectancy before and after treatment are interrelated, it was not possible to include all in the same analysis.

All analyses were performed using R, version 4.0.2 [27].

Results

Sample

Data was collected between the 9th of April and the 5th of June 2020. In total, 3,387 participants provided informed consent. Of these participants, 1,143 (33.7%) completed the questionnaire. People who started but did not complete the questionnaire were on average older (50.4 versus 43.7 years), more often female (66% versus 56%) and more often had a low educational level (35% versus 25%) than people who did complete the questionnaire.

Table 2 displays the age, gender and educational level of the sample. It shows that, compared to the Dutch general population, in our sample, people older than 65 years of age, people with a low educational level and males were slightly underrepresented.

Table 2: Descriptive characteristics of the sample

		Study sample		**Dutch general population**[b]
		N (%)	Mean (SD[a])	%
Age			43.7 (16.1)	
	18-19 years	48 (4.2)		3.1
	20-39 years	457 (40.0)		31.1
	40-64 years	519 (45.4)		41.6
	65-79 years	116 (10.1)		18.2
	> 79 years	3 (0.3)		6.0
Gender	Males	496 (43.4)		49.7
	Females	643 (56.3)		50.4
	Other	4 (0.3)		-
Educational level[c]	Low	280 (24.5)		30.2
	Middle	424 (37.1)		36.8
	High	434 (38.0)		31.5
	Missing/Don’t know	5 (0.4)		-

[a] SD= standard deviation

[b] Source: CBS Statline: https://opendata.cbs.nl/statline/#/CBS/en/

[c] Education levels correspond to the SOI 2016 and the ISCED 2011 classifications

The answers to the questions concerning how participants experienced the PVE are included in Appendix D.

Main analysis

The results of the portfolio choice model are presented in Table 3. These results show that age of the patient group, gain in remaining life expectancy, gain in quality of life, and savings in addition to the imposed minimum savings of €100 million have a statistically significant effect on the likelihood of disinvesting a treatment. The larger the health gain of the treatment, the less likely participants are to disinvest the treatment. From the coefficients, it can be calculated that a gain in life expectancy of one year provides similar utility to participants as a gain in quality of life of 3%. The negative sign of the additional savings coefficient indicates that participants prefer not to save more than the minimum required savings of €100 million.

As previous studies found the preference for age of the patient to have an inverse U-shaped pattern [17], we included age of the patient squared to check whether this was also the case in our study. The statistical significance of this attributes may indicate that the preference for age of the patient group in this study may also have a U-shaped pattern.

To illustrate the findings in Table 3, we have calculated the relative utility lost by disinvesting three randomly selected portfolios that participants could compile in version 1 of the design of our PVE and that meet the required minimum savings of €100 million. The results of these calculations can be found in Appendix E. Of the three portfolios in Appendix E, participants would choose to disinvest portfolio 1 as disinvesting this portfolio results in the smallest loss in utility.

Table 3: Portfolio choice model

Attribute	Levels	Coefficient (SE[a])
Age of the patient (per 10 years)		0.0417 (0.0283)
Age of the patient squared (per 10 years)		-0.0107** (0.0038)
Gain in remaining life expectancy (per year)		0.0615*** (0.0041)
Gain in quality of life (per 1%)		0.0214*** (0.0016)
Alternative treatment (reference category = no alternative treatment)	There is an alternative treatment available that only treats symptoms	0.0272 (0.0508)
	There is an alternative treatment available that treats the disease	-0.0678 (0.0510)
Marginal utility of additional savings (per 1 million Euros)		-0.0104*** (0.0010)
Log-Likelihood		-5168
AIC		10351
BIC		10386
Observations		1143

[a] Standard error

***p < 0.001, **p < 0.01, *p < 0.05

Quality of life and life expectancy before and after treatment

The results of the analyses to assess the association between the preference for the quality of life and life expectancy before and after treatment and the preference for the gain in quality of life and life expectancy are displayed in Tables 4 and 5. Table 4 shows that both a larger gain in remaining life expectancy and a larger remaining life expectancy before treatment increase the likelihood to maintain the reimbursement of the treatment. However, the negative coefficient of the interaction effect indicates that if the remaining life expectancy before treatment is higher, people attach less value to the same gain in life expectancy. Quality of life before treatment does not have a statistically significant effect on the likelihood to disinvest a treatment.

Table 4: Portfolio model including quality of life and remaining life expectancy before treatment

Attribute	**Levels**	**Coefficient (SE[a])**
Age of the patient (per 10 years)		0.0076 (0.0381)
Age of the patient squared (per 10 years)		-0.0077 (0.0047)
Gain in remaining life expectancy (years)		0.0979*** (0.0113)
Gain in quality of life (1%)		0.0120* (0.0059)
Remaining life expectancy before treatment (years)		0.0105** (0.0047)
Quality of life before treatment (1%)		-0.0008 (0.0015)
Gain in life expectancy × Life expectancy before		-0.0030*** (0.0008)
Gain in quality of life × Quality of life before		0.0002 (0.0001)
Alternative treatment (reference category = no alternative treatment)	There is an alternative treatment available that only treats symptoms	0.0216 (0.0536)
	There is an alternative treatment available that treats the disease	-0.0833 (0.0541)
Marginal utility of additional savings (per 1 million Euros)		-0.0099*** (0.0010)
Log-Likelihood		-5160
AIC		10341
BIC		10397
Observations		1143

[a] Standard error

***p < 0.001, **p < 0.01, *p < 0.05

Table 5 shows that the value attached to the same gain in remaining life expectancy is smaller if the remaining life expectancy after treatment is higher. Table 5 also shows that if the quality of life after treatment is better, that the value attached to the same gain in quality of life is smaller. The utility lost by disinvesting the three randomly selected portfolio's in Appendix E was also calculated based on the additional analyses. The results of these calculations are displayed in Appendix F.

Table 5: Portfolio model including quality of life and remaining life expectancy after treatment

Attribute	**Levels**	**Coefficient (SE[a])**
Age of the patient (per 10 years)		-0.0339 (0.0387)
Age of the patient squared (per 10 years)		-0.0036 (0.0048)
Gain in remaining life expectancy (years)		0.1030*** (0.0128)
Gain in quality of life (1%)		0.0493*** (0.0113)
Remaining life expectancy after treatment (years)		0.0040 (0.0039)
Quality of life after treatment (1%)		0.0023 (0.0012)
Gain in life expectancy × Life expectancy after		-0.0018*** (0.0005)
Gain in quality of life × Quality of life after		-0.0004** (0.0001)
Alternative treatment (reference category = no alternative treatment)	There is an alternative treatment available that only treats symptoms	-0.0284 (0.0531)
	There is an alternative treatment available that treats the disease	-0.1112* (0.0535)
Marginal utility of additional savings (per 1 million Euros)		-0.0096*** (0.0010)
Log-Likelihood		-5154.2674
AIC		10330.5348
BIC		10385.9903
Observations		1143

[a] Standard error
***p < 0.001, **p < 0.01, *p < 0.05

Discussion

The main objective of this study was to investigate the preferences of Dutch citizens towards the relative importance of attributes of healthcare interventions in the context of disinvestment. We found that participants prefer to disinvest treatments with limited to no gain in quality of life and life expectancy and that are targeted at older patient groups. Furthermore, we found that participants prefer to disinvest portfolio's (i.e. combinations of healthcare interventions) with smaller additional savings (i.e. savings in addition to the minimum required savings) over portfolio's with larger additional savings. This implies that they aimed to save no more than the minimum required savings of €100 million. As we told participants that additional savings would be spent elsewhere in the healthcare system, this finding may indicate that participants value the maintained treatments over other means in healthcare the money could be spent on. On the other hand, it may also indicate that people did not want to save more than the minimum required savings because they already considered this amount too high, or it may reflect a general dislike of the disinvestment task.

The secondary objective of this study was to assess what the effect of the quality of life and life expectancy before and after treatment is on the importance attached by Dutch citizens to a gain in quality of life and life expectancy in disinvestment decisions. We found that the value attached to a gain in quality of life is affected by quality of life after treatment: if the quality of life after treatment is better, less value is attached to the same gain in quality of life, compared to when quality of life after treatment is worse. Furthermore, we also found that the value attached to a gain in remaining life expectancy is affected by the remaining life expectancy before as well as after treatment. The model results showed that the value attached to such a gain is smaller if life expectancy before/after treatment is higher.

Comparison with literature

Comparison with disinvestment literature

Our results confirm the findings of two previous studies on disinvestment in which effectiveness of the treatment was considered an important attribute [18-20]. Street et al. found that this attribute was rarely considered in isolation from other attributes [20], which is also confirmed by the findings of our study. For instance, just as Street et al., we found that participants trade the gain in health against disease severity/health status. However, contrary to the previous literature [18, 20], the availability of an alternative treatment did not significantly affect the likelihood to disinvest a treatment. Hence, the other attributes were clearly considered to be more important than availability of alternative treatment in disinvestment decision-making.

Comparison with the investment literature

Our finding that health gain is an important attribute for disinvestment, corresponds with the findings of two systematic reviews on allocation preferences in the investment context [16, 17]. These reviews found that health gain is consistently highly valued in previous studies and can, therefore, be considered one of the most important attributes. As in this study, the reviews found that larger health gains are preferred over smaller health gains, but that these gains are weighted against disease severity and age of the patient group [16, 17].

Hence, the results of this study are very much aligned with the findings of preference studies in the investment context. This may indicate that, with regards to important attributes, the disinvestment context may not be that different from the investment context after all. A possible explanation for this may be the lack of information provided on the exact treatments and patient groups concerned. As this information was not available, the patients who would potentially be affected by disinvestment were not identifiable, which may have made the task more abstract to participants, making it a more rational trade-off than if the patients potentially affected by disinvestment would have been identifiable [28-30]. Hence, more differences between the investment and disinvestment context may have been found if the patient group concerned would have been known. We recommend future research to explore whether this hypothesis is true.

Reflections on the participatory value evaluation method

PVE is a novel stated preference method that resembles other stated preference methods such as Discrete Choice Experiments (DCE). To our knowledge, this study is the first PVE in the context of the allocation of healthcare budgets. However, numerous DCEs have already been conducted on this topic [31-36]. There are two main differences between DCE and PVE. First, in DCE, participants express their preferences through selecting a *single* healthcare intervention out of two or more healthcare interventions, whilst in PVE, participants select a *bundle* of healthcare interventions out of a larger bundle of healthcare interventions [25]. Second, in PVE, the total societal budget impact is part of the experiment, while in DCE this is not the case. By incorporating this budget in the experiment, participants can express their preferences towards (the attributes of) specific healthcare interventions as well as the extent to which scarce healthcare resources should be allocated to these healthcare interventions [25]. Because of these two differences, PVE more closely aligns with the practice of societal decision-making than DCE does. This close alignment to the practice of policymaking allowed us to involve citizens in the complexity of making decisions on the disinvestment of healthcare interventions. Nevertheless, as DCE requires participants to make multiple choices, DCE may be more efficient in obtaining information on the relative importance of attributes. Therefore, when deciding between PVE and DCE to measure public preferences, researchers should determine whether they are interested in 1) involving participants in the complexity of societal decision-making, 2) the amount of budget participants want to (re-)allocate, or 3) efficiently measuring the preferences for a number of attributes. In the former two situations, PVE may be more suitable, while in the latter situation, DCE may be more suitable.

In this study, we used a non-labelled PVE approach. Alternatively, a labelled approach may have been used in which the candidate interventions for disinvestment would be given identifying labels such as “statins”, “contraceptives”, and “cognitive behavioural therapy”. An advantage of this labelled approach is that it more closely aligns with the practice of disinvestment decision-making, as in that case, the exact intervention and patient group is also known. Hence, in a labelled approach, the differences between DCE and PVE are more distinct, and, as such, the advantages of using PVE are clearer. However, we anticipated that if we would have labelled the treatments, these labels may have dominated the preferences participants have. This is something that has also been observed in PVE’s on infrastructure projects [24]. A consequence of this is that we would not be able to assess the importance of the different attributes of healthcare interventions in disinvestment decisions, which was the main aim of this study. Moreover, another advantage of the non-labelled approach is that the findings are applicable to a broad range of disinvestment decisions, instead of only the labelled cases included in a labelled PVE. Nevertheless, in transferring the findings of this study to the practice of disinvestment decision-making, policymakers are recommended to also take the context of disinvestment into account as previous research has shown that contextual aspects, such as view on the disinvestment case, support and financial incentives, are very important in disinvestment decision-making [14].

Context

The data for this study were collected at the time the first wave of the Covid-19 epidemic took place in the Netherlands, which resulted in tremendous pressure on the Dutch healthcare system. Regular healthcare was delayed, and it was feared that the intensive care capacity in the Netherlands would be insufficient [37]. Healthcare providers were under much pressure, working long hours. Their efforts were very much appreciated by Dutch citizens. At the same time, the country was in partial lock-down with schools, bars and many shops being closed, and citizens being asked to work from home. Hence, data was collected in exceptional circumstances. This may have affected the findings of this PVE. For instance, because of the large appreciation of Dutch citizens for healthcare providers they may either have been more reluctant (e.g. consider it important to let healthcare providers determine what is valuable care) or less reluctant to disinvest treatments (e.g. to get some weight of the shoulders of healthcare providers). To limit the effect of this unique situation on the findings of our study, we specifically told participants that acute healthcare, such as healthcare targeted at people suffering from infectious diseases like Covid-19 would never be disinvested. Despite this, our findings may still be affected by the exceptional circumstances.

Strengths and limitations

Because of the structured, evidence-base selection of attributes and attribute levels, this study can be compared with previous studies using other methods to assess healthcare allocation preferences. Moreover, by including both remaining life expectancy as well as age, we were able to disentangle the preferences for life expectancy from the preferences for age [16, 17]. Furthermore, the face-to-face pilot test enabled us to assess and increase the feasibility of the PVE method.

Nevertheless, this study also has limitations. First, drop-out was quite high with only a third of participants who started the questionnaire also completing it. Obviously, the task given was not a popular one, no one likes to think about necessary savings on healthcare. Moreover, the task was cognitively demanding, as participants had to compare characteristics of eight different imaginary treatments in one single task. As discussed in the results section, drop-out may be selective with participants dropping-out being on average older, lower educated and more often female. Moreover, drop-out may also be selective in the sense that only people who were most motivated completed the PVE-task. This may be especially problematic if participants completing the PVE-task have a different perspective on disinvestment than non-completers have. It is not possible to assess whether this is the case, as only descriptive information of participants who dropped-out was available. However, it is important to take this into account when interpreting the results of this study. Second, this study has been conducted in the Dutch context. As it is clear that preferences may be time and context-specific, researchers are recommended to take the context into account when transferring these results to their own setting.

Policy implications

The results of this study have implications for disinvestment decision-making. First, the model results of this study show that citizens prefer to disinvest treatments that result in limited or no health gain and that are targeted at older patient groups. This information can be used in the selection of candidate interventions for disinvestment. Furthermore, it stresses the need to de-implement low-value care (i.e. healthcare interventions that provide little to no clinical benefit [38]), as has also been suggested by international de-implementation campaigns such as Choosing Wisely [38, 39]. In addition, this information can be used in the communication about disinvestment decisions. Based on the results of this study, we expect that if policymakers explain to the public that a disinvested treatment does not have any or only limited health effect, this will increase societal support.

Second, the PVE method shows that subgroups of the general public are able to make disinvestment decisions. This indicates that it is feasible to involve this subgroup of citizens in disinvestment processes. For instance, their advice can be asked on the prioritization of disinvestment candidates. However, the strong dislike of disinvestment of pilot test participants, the large number of selective drop-outs and the feedback provided by participants showed that there was also a significant subgroup of the general public who are not able or dislike making disinvestment decisions to such a degree that they will not be willing/able to advice policymakers on these difficult decisions. This implies that additional efforts are needed to also fully engage these subgroups of the Dutch population in disinvestment decisions.

Finally, responses to the follow-up questions in this PVE, displayed in Appendix D, showed that the majority of participants feel that the advice of experts deserves more weight than the advice of citizens with regards to disinvestment decisions. Nevertheless, 92% of participants who answered the question (72% of total) feel that the advice of citizens should be taken into account as well. Therefore, policymakers are recommended to think of ways to incorporate the preferences of citizens in disinvestment decision-making, in addition to the advice of experts. The majority of participants (58% of those who answered this question, 50% of total) suggest that PVE may be a good method to involve citizens in this type of complicated and sensitive policy decisions. However, as discussed in a previous paragraph, this method may only be suitable for a part of the general population or requires additional efforts to engage a representative sample of the general population. Therefore, it may also be valuable to explore whether other methods (e.g. citizen juries or PVE's with researcher assistance) are better suitable to involve a representative sample of the general population in disinvestment decision-making.

Conclusions and recommendations

In this study, we found that citizens prefer to disinvest treatments with limited to no health gain and that are targeted at older patient groups. The weight attached to the gain in health depends on the health status before and/or after treatment. Policymakers are recommended to take these preferences into account in the selection of candidate interventions for disinvestment as well as in the communication on disinvestment decisions to increase the support for these decisions.

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Appendices

Appendix A: Survey instruction

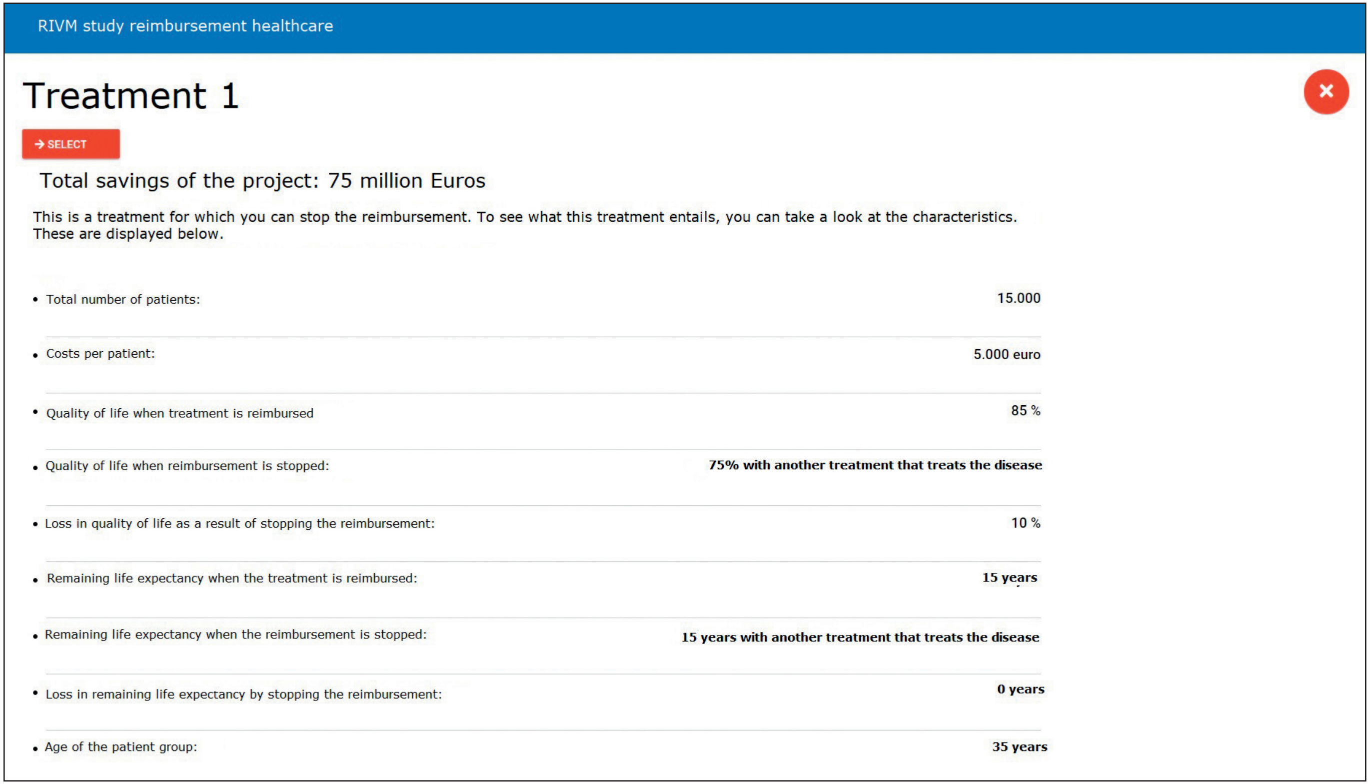
Section	Webpage	Text
Introduction	Introduction	This research is about the costs of healthcare. In the Netherlands, healthcare costs 5,500 euros on average per inhabitant per year. These costs are still rising every year. The government is asking citizens to think along about cutbacks in health care. What could be removed from the basic benefit package, in your opinion? And which care would you definitely want to keep included? We are going to show you 8 different treatments. Your task is to remove one or more treatments from the basic benefit package. Is it really necessary to make cuts? For this exercise, it is. Some treatments will have to be removed from the basic benefit package because many new treatments are going to be added to the basic benefit package. New medicines, for example, or new ways to diagnose and treat diseases. To prevent health insurance premiums rising much more, it is also necessary to remove treatments from the basic benefit package from time to time. Please be assured that acute care, for example, infectious disease control such as Covid-19 control or care for people with a heart attack, will always be reimbursed.
Instruction	Attributes and levels	Each treatment on the list is described using a number of characteristics. These characteristics are explained on this page. **Number of patients in the Netherlands** Some diseases only occur in very few patients. Other diseases are very common. Here you can see how many people are currently receiving the treatment in the Netherlands. At the moment, all these patients are reimbursed for this treatment. **Cost per patient** This is the cost of the treatment per patient. When we talk about costs, we mean all the costs of care taken together, such as the costs of medicines, GPs and hospital costs. Some treatments are cheap, for example, 100 euros per patient. Other treatments are very expensive. Some treatments may cost as much as 50,000 or 90,000 euros per patient. **Quality of life when the treatment is reimbursed** This is the quality of life of patients when the treatment is reimbursed. Here you can see patients' average quality of life with the treatment. We indicate quality of life with a number between 0% and 100%. 100% represents the best quality of life you can imagine. 0% represents the worst quality of life you can imagine.

Section	Webpage	Text
		Quality of life if reimbursement for the treatment is discontinued. This is the quality of life of patients if reimbursement of the treatment is discontinued. We indicate quality of life with a number between 0% and 100%. 100% represents the best quality of life you can imagine. 0% represents the worst quality of life you can imagine. If reimbursement for treatment is discontinued, there are three options: - No other treatment is available. The number represents the quality of life without the treatment. - The only treatment available is to alleviate symptoms of the disease. This treatment is still reimbursed. The number represents the quality of life with this other treatment. - A different treatment is available. This treatment is still reimbursed. The number represents the quality of life with this other treatment. **Loss of quality of life due to discontinuation of reimbursement** Here you can see the loss of quality of life of patients due to discontinuation of the reimbursement of the treatment. Sometimes, discontinuing reimbursement for treatment will not change the quality of life. For example, because a suitable alternative treatment is available or because the treatment has no effect on the quality of life. If this is the case, you will see the value 0%. **Remaining life expectancy when reimbursed for the treatment** This is the remaining life expectancy of patients when the treatment is reimbursed. Here you can read how many years on average patients stay alive with the treatment. **Remaining life expectancy in case of discontinuation of the treatment** This is the remaining life expectancy of patients when reimbursement of the treatment is discontinued. Here you can see how many years on average patients will live when the reimbursement of the treatment is discontinued. If reimbursement for treatment is discontinued, there are three options: - No other treatment is available. The number represents the remaining life expectancy without the treatment. - The only treatment available is to alleviate symptoms of the disease. This treatment is still reimbursed. The number represents the remaining life expectancy of patients with this other treatment. - A different treatment is available. This treatment is still reimbursed. The number represents the remaining life expectancy of patients with this other treatment.

Section	Webpage	Text
		Reduction in life expectancy due to discontinuation of reimbursement Here you can see the reduction in life expectancy of patients due to discontinuation of reimbursement of the treatment. Sometimes, discontinuing reimbursement of the treatment will not change life expectancy. For example, because a suitable alternative treatment is available or because the treatment does not affect life expectancy. If this is the case, you will see the value 0 years. **Age of the patient** This is the average age of the patients who are eligible for the treatment.
	Introduction webtool	On the main screen, you can click on all 8 treatments for more information. The 8 treatments are described using a number of characteristics. The treatments differ only for these characteristics. In other words, you can assume that the treatments do not differ in other characteristics, such as side effects. You may be wondering why we talk about ‘treatment’ and don’t state exactly what treatment for what disease this is. We do this because we want you to look closely at the characteristics of the treatment rather than choose based on the treatment name. We ask you to save at least 100 million euros by choosing a few treatments that can be removed from the basic benefit package. It is fine if you save more than 100 million euros. You can assume that in that case, fewer cutbacks will be needed in the future.
	Instruction options webtool	Please see below for an instruction video. This video is 3 minutes long and can be viewed on a large screen. Please start by watching this video. You can also read the instructions again below. These instructions contain the same information as the instruction video. **Information about treatment** There is an information button next to every treatment; the ‘info’ button. If you click on the information button, you will see information about the treatment. We recommend that you read the information for all eight treatments. If you move your mouse over a characteristic, you will be shown more information about what a characteristic means. To return to the main menu, click on the red button with the white cross. In the main menu, to the left of the treatment, in the grey circles, you can see again how much money you will save if you discontinue reimbursement of the treatment. If you are already sure that you want to discontinue reimbursement of a treatment, you can indicate this with the ‘select’ button in the main menu and on the treatment information page.

Section	**Webpage**	**Text**
		Comparison Once you have indicated which treatments you want to compare, click on the red ‘compare’ button at the top of the page. You can also click on the green button with the black arrow on the left of the page. To return to the main menu, click on the ‘back’ button. You can also arrange the treatments according to the various characteristics. To do so, click on “choose a characteristic” behind “sort by” at the top centre of the screen. You can now select a characteristic here to rank on. The score of the treatments for this characteristic is placed behind the treatments. The treatments are ranked in order of how they score on the feature. **Selection** To select a treatment, you can click on the toggle under ‘selection’. The treatment will then appear on the right side of the screen. At the top right of the page, you can see that you need to make cuts of 100 million euros. Below it, you can see the total savings of all the treatments you have chosen. Below that number, you can see how much you still need to save. **Confirm selection** Once you have reached cutbacks of 100 million or more, you can review your selection by clicking on the red ‘selection overview’ button at the top of the screen. If you are sure of your choice, you can confirm your choice here by clicking on the “confirm” button. If you still want to make a change, you can go back to the selection screen using the “back” button. We recommend that you do NOT use the refresh button of your browser while you complete the form. If you click refresh, you will go back to the start of the questionnaire and have to start over. After you have made your choice, we will ask you a few more questions.

Appendix B: Visual presentation of web-based environment

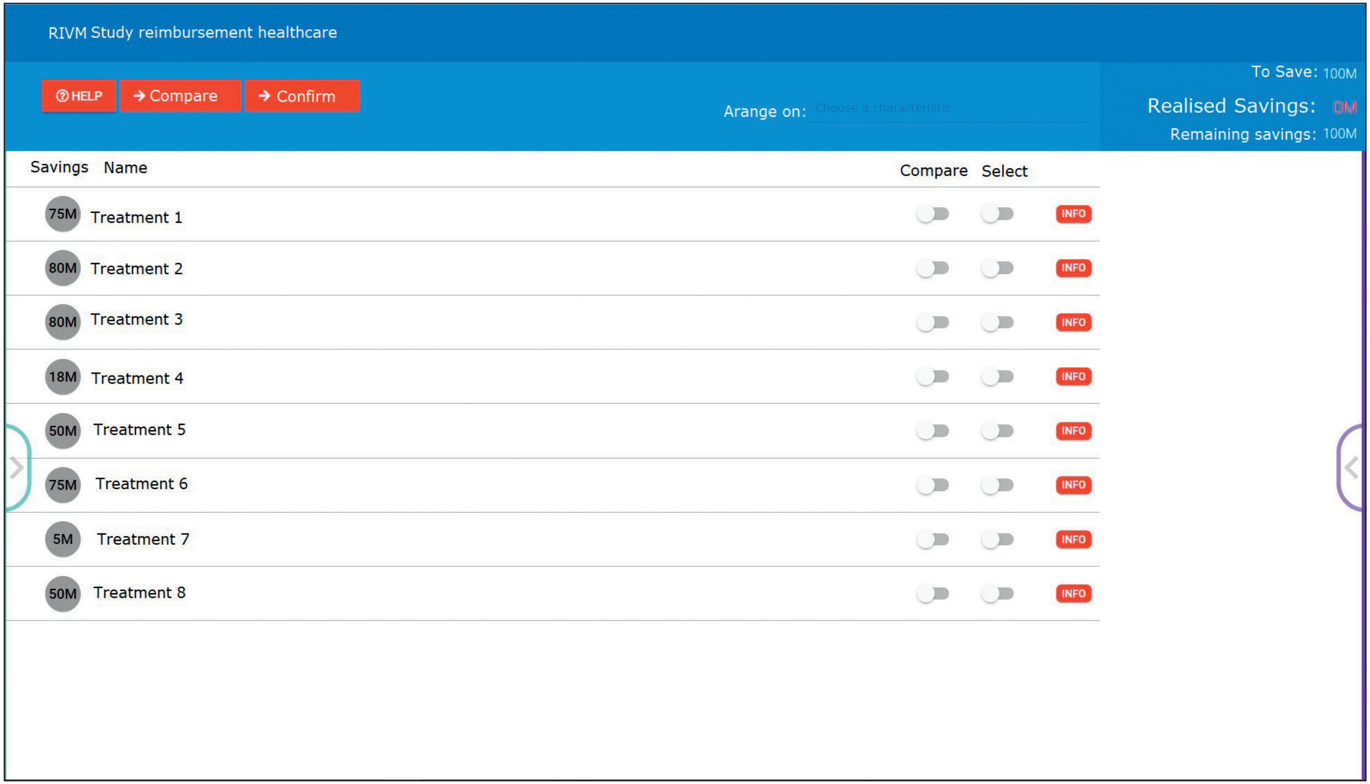


Treatment 1

Total savings of the project: 75 million Euros

This is a treatment for which you can stop the reimbursement. To see what this treatment entails, you can take a look at the characteristics. These are displayed below.

• Total number of patients:	15.000
• Costs per patient:	5.000 euro
• Quality of life when treatment is reimbursed	85 %
• Quality of life when reimbursement is stopped:	**75% with another treatment that treats the disease**
• Loss in quality of life as a result of stopping the reimbursement:	10 %
• Remaining life expectancy when the treatment is reimbursed:	**15 years**
• Remaining life expectancy when the reimbursement is stopped:	**15 years with another treatment that treats the disease**
• Loss in remaining life expectancy by stopping the reimbursement:	**0 years**
• Age of the patient group:	**35 years**



To Save: 100M

Realised Savings: 0M

Remaining savings: 100M

Savings	Name	Compare	Select
75M	Treatment 1	○	○
80M	Treatment 2	○	○
80M	Treatment 3	○	○
18M	Treatment 4	○	○
50M	Treatment 5	○	○
75M	Treatment 6	○	○
5M	Treatment 7	○	○
50M	Treatment 8	○	○

Appendix C: Attribute level selection

The levels for the ‘availability of alternative treatment’ attribute were obtained from Bourke et al. 2018 [31]. The levels for ‘average age of the patient group’ were adapted from van de Wetering et al. 2016 [35] for the purpose of this study. The levels for the health effect attributes were determined based on the authors’ knowledge of previous reimbursement decisions in the Netherlands combined with input from the literature.

The budget-impact levels were restricted between 1 and 90 million Euros and calculated by multiplying levels of ‘number of patients’ and ‘costs per patient’. 90 Million Euros was chosen as an upper bound to force participants to select at least 2 treatments for disinvestment (note that the minimum budget to be saved was 100 million Euros). The lower bound of 1 million Euros was based on a trade-off between impact of the budget-impact on the total budget to be saved, and feasibility for the levels of the underlying attributes. The levels for ‘number of patients’ and ‘costs per patient’ were determined based on the following considerations: 1) a maximum of six levels for each attribute, 2) including a broad range, from cheap treatments for many people to expensive treatments for just a few patients, 3) the possibility to make combinations between the ‘number of patients’ and the ‘costs per patient’, resulting in levels of the budget-impact within the restrictions posed, 4) the possibility to combine each level of ‘number of patients’ with at least 2 levels of ‘costs per patient’ and vice versa, 5) sufficient overlap between the combinations of the levels of ‘number of patients’ and ‘costs per patient’. Considerations 1, 4 and 5 were included to ensure efficiency of the design. Consideration 2 was included to keep the design realistic.

Appendix D: Experience participants with PVE task

Question	Answer (N (%))					
	Completely agree	**Agree**	**Don't agree, don't disagree**	**Disagree**	**Completely disagree**	**Missing**
I had enough information to make a choice	103 (9)	460 (40)	254 (22)	134 (12)	44 (4)	148 (13)
The costs of the different treatments were decisive in making my choice	93 (8)	345 (30)	298 (26)	189 (17)	68 (6)	150 (13)
I am convinced of my choice	90 (8)	389 (34)	349 (34)	119 (10)	44 (4)	152 (13)
I consider this to be a realistic study	75 (7)	330 (29)	340 (30)	176 (15)	68 (6)	154 (13)
I found it difficult to understand the task	39 (3)	138 (12)	218 (19)	354 (31)	240 (21)	154 (13)
I found it difficult to make a choice	181 (16)	365 (32)	209 (18)	176 (15)	59 (5)	153 (13)
By participating in this experiment, I learned more about the choices the government needs to make regarding the reimbursement of healthcare	110 (10)	479 (42)	259 (23)	103 (9)	45 (4)	147 (13)
By participating in this experiment, I learned more about the choices the government needs to make to keep healthcare affordable	97 (8)	503 (44)	257 (22)	97 (8)	39 (3)	150 (13)
This is a good method to involve citizens in reimbursement decisions	150 (13)	428 (37)	233 (20)	113 (10)	69 (6)	150 (13)
The government should use this method more often to involve citizens in government policy	168 (15)	416 (36)	233 (20)	108 (9)	69 (6)	149 (13)
Because of the involvement of citizens in decision, the final government decision is more acceptable to me	123 (11)	414 (36)	263 (23)	122 (11)	62 (5)	159 (14)

	Government should follow the advice of (N (%)):					
	Citizens	Citizens and experts, with more weight to citizens	Citizens and experts, with equal weight	Experts and citizens, with more weight to experts	Experts	No Answer
We also consulted a number of scientists who are expert in the field of the reimbursement of healthcare. How much value should the government attach to the advice of citizens compared to the advice of experts	47 (4)	85 (7)	255 (22)	441 (39)	77 (7)	238 (21)

Appendix E: Relative utility lost by disinvesting three different portfolios of treatments

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treatments	**Attributes**	**Attribute levels**	**Utility**	**Chosen treatments**	**Attributes**	**Attribute levels**	**Utility**	**Chosen treatments**	**Attributes**	**Attribute levels**	**Utility**
Treatment 4	Costs	20M		Treatment 1	Costs	80M		Treatment 1	Costs	80M	
	Gain in quality of life	5%	0.1070		Gain in quality of life	45%	0.9630		Gain in quality of life	45%	0.9630
	Gain in life expectancy	1 year	0.0615		Gain in life expectancy	5 years	0.3075		Gain in life expectancy	5 years	0.3075
	Age	35 years	0.0149		Age	10 years	0.0310		Age	10 years	0.0310
	Alternative treatment	Yes, treats symptoms	0.0272		Alternative treatment	Yes, treats disease	-0.0678		Alternative treatment	Yes, treats disease	-0.0678
	Total		**0.2106**		Total		**1.2337**		Total		**1.2337**
Treatment 5	Costs	20M		Treatment 3	Costs	1M		Treatment 2	Costs	10M	
	Gain in quality of life	5%	0.1070		Gain in quality of life	25%	0.5350		Gain in quality of life	25%	0.5350
	Gain in life expectancy	0 years	0		Gain in life expectancy	5 years	0.3075		Gain in life expectancy	10 years	0.6150
	Age	10 years	0.0310		Age	55 years	-0.0943		Age	55 years	-0.0943
	Alternative treatment	Yes, treats disease	-0.0678		Alternative treatment	Yes, treats disease	-0.0678		Alternative treatment	Yes, treats symptoms	0.0272
	Total		**0.0702**		Total		**0.6804**		Total		**1.0829**

Portfolio 1				**Portfolio 2**				**Portfolio 3**			
Chosen treatments	**Attributes**	**Attribute levels**	**Utility**	**Chosen treatments**	**Attributes**	**Attribute levels**	**Utility**	**Chosen treatments**	**Attributes**	**Attribute levels**	**Utility**
Treatment 6	Costs	40M		Treatment 4	Costs	20M		Treatment 8	Costs	20M	
	Gain in quality of life	0%	0		Gain in quality of life	5%	0.1070		Gain in quality of life	5%	0.1070
	Gain in life expectancy	0.5 years	0.0308		Gain in life expectancy	1 year	0.0615		Gain in life expectancy	5 years	0.3075
	Age	55 years	-0.0943		Age	35 years	0.0149		Age	75 years	-0.2891
	Alternative treatment	No	0		Alternative treatment	Yes, treats symptoms	0.0272		Alternative treatment	No	0
	Total		**-0.0636**		Total		**0.2106**		Total		**0.1254**
Treatment 8	Costs	20M		Treatment 7	Costs	2M					
	Gain in quality of life	5%	0.1070		Gain in quality of life	5%	0.1070				
	Gain in life expectancy	5 years	0.3075		Gain in life expectancy	15 years	0.9225				
	Age	75 years	-0.2891		Age	35 years	0.0149				
	Alternative treatment	No	0		Alternative treatment	Yes, treats symptoms					
	Total		**0.1254**		Total		**1.0716**				

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treat-ments	**Attributes**	**Attribute levels**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute levels**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute levels**	**Utility**
Subtotal utility lost			**0.3426**	**Subtotal utility lost**			**3.1962**	**Subtotal utility lost**			**2.4420**
Additional savings (in addition to the 100M)		0	0	Additional savings (in addition to the 100M)		3M	-0.0312	Additional savings (in addition to the 100M)		10M	-0.1040
Total utility lost by disinvesting portfolio[a]			**0.3426**	**Total utility lost by disinvesting portfolio**[a]			**3.2274**	**Total utility lost by disinvesting portfolio**[a]			**2.5460**

[a] The total utility lost by choosing the portfolio for disinvestment: $U_{n\bar{p}} = \sum_{j=1}^{J}\left(1 - y_{nj}\right) \cdot U_{nj} - \alpha\left(\sum_{j=1}^{J}\left(1 - y_{nj}\right) \cdot c_{nj} - B\right) + \varepsilon_{np}$

Appendix F: Relative utility lost by disinvesting three different portfolios based on the additional analysis including interactions between gain in health and health status before and after treatment

Table F1: Relative utility lost by disinvesting three different portfolios of treatments based on the portfolio model including the health status before treatment

Portfolio 1				**Portfolio 2**				**Portfolio 3**			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 4	Costs	20M		Treatment 1	Costs	80M		Treatment 1	Costs	80M	
	Gain in quality of life	5%	0.0750		Gain in quality of life	45%	0.8270		Gain in quality of life	45%	0.8270
	Quality of life before	75%			Quality of life before	35%			Quality of life before	35%	
	Gain in life expectancy	1 year	0.1354		Gain in life expectancy	5 years	0.3770		Gain in life expectancy	5 years	0.3770
	Life ex-pectancy before	5 years			Life ex-pectancy before	25 years			Life ex-pectancy before	25 years	
	Age	35 years	-0.0677		Age	10 years	-0.0001		Age	10 years	-0.0001
	Alternative treatment	Yes, treats symptoms	0.0216		Alternative treatment	Yes, treats disease	-0.0833		Alternative treatment	Yes, treats disease	-0.0833
	Total		**0.1643**		Total		**1.1206**		Total utility		**1.1206**

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 5	Costs	20M		Treatment 3	Costs	1M		Treatment 2	Costs	10M	
	Gain in quality of life	5%	0.0750		Gain in quality of life	25%	0.5310		Gain in quality of life	25%	0.5310
	Quality of life before	75%			Quality of life before	55%			Quality of life before	55%	
	Gain in life expectancy	0 years	0.1575		Gain in life expectancy	5 years	0.3770		Gain in life expectancy	10 years	0.8815
	Life ex-pectancy before	15 years			Life ex-pectancy before	25 years			Life ex-pectancy before	5 years	
	Age of the patient group	10 years	-0.0001		Age of the patient group	55 years	-0.1911		Age of the patient group	55 years	-0.1911
	Alternative treatment	Yes, treats disease	-0.0833		Alternative treatment	Yes, treats disease	-0.0833		Alternative treatment	Yes, treats symptoms	0.0216
	Total		**0.1491**		Total		**0.6336**		Total		**1.2430**

Portfolio 1				**Portfolio 2**				**Portfolio 3**			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 6	Costs	40M		Treatment 4	Costs	20M		Treatment 8	Costs	20M	
	Gain in quality of life	0%	-0.0440		Gain in quality of life	5%	0.0750		Gain in quality of life	5%	0.0750
	Quality of life before	55%			Quality of life before	75%			Quality of life before	75%	
	Gain in life expectancy	0.5 years			Gain in life expectancy	1 years			Gain in life expectancy	5 years	
	Life ex-pectancy before	1 years	0.0580		Life ex-pectancy before	5 years	0.1354		Life ex-pectancy before	1 year	0.4850
	Age of the patient group	55 years	-0.19113		Age of the patient group	35 years	-0.0677		Age of the patient group	75 years	-0.3761
	Alternative treatment	No	0		Alternative treatment	Yes, treats symptoms	0.0216		Alternative treatment	No	0
	Total		**-0.1771**		Total		**0.1643**		Total		**0.1839**

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 8	Costs	20M		Treatment 7	Costs	2M					
	Gain in quality of life	5%	0.0750		Gain in quality of life	5%	0.0750				
	Quality of life before	75%			Quality of life before	75%					
	Gain in life expectancy	5 years	0.4850		Gain in life expectancy	15 years	0.9510				
	Life ex-pectancy before	1 year			Life ex-pectancy before	15 years					
	Age of the patient group	75 years	-0.3761		Age of the patient group	35 years	-0.0677				
	Alternative treatment	No	0		Alternative treatment	Yes, treats symptoms	0.0216				
	Total		**0.1839**		Total		**0.9799**				
Additional savings (in addition to the 100M)		0	0	Additional savings (in addition to the 100M)		3M	-0.0297	Additional savings (in addition to the 100M)		10M	-0.0990
Total utility lost by disinvesting portfolio			**0.3201**	**Total utility lost by disinvesting portfolio**			**2.9280**	**Total utility lost by disinvesting portfolio**			**2.6465**

Table F2: Relative utility lost by disinvesting three different portfolios of treatments based on the portfolio model including the health status after treatment

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treatments	**Attributes**	**Attribute level**	**Utility**	**Chosen treatments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treatments**	**Attributes**	**Attribute level**	**Utility**
Treatment 4	Total costs	20M		Treatment 1	Total costs	80M		Treatment 1	Total costs	80M	
	Gain in quality of life	5%	0.2705		Gain in quality of life	45%	0.9625		Gain in quality of life	45%	0.9625
	Quality of life after	80%			Quality of life after	80%			Quality of life after	80%	
	Gain in life expectancy	1 year	0.1162		Gain in life expectancy	5 years	0.3650		Gain in life expectancy	5 years	0.3650
	Life expectancy after	6 years			Life expectancy after	30 years			Life expectancy after	30 years	
	Age of the patient group	35 years	-0.1628		Age of the patient group	10 years	-0.0375		Age of the patient group	10 years	-0.0375
	Alternative treatment	Yes, treats symptoms	-0.0284		Alternative treatment	Yes, treats disease	-0.1112		Alternative treatment	Yes, treats disease	-0.1112
	Total utility treatment		**0.1956**		Total utility		**1.1788**		Total utility		**1.1788**

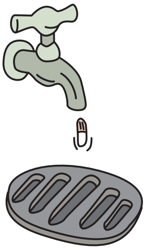
Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 5	Total costs	20M	0.2705	Treatment 3	Total costs	1M	0.6165	Treatment 2	Total costs	10M	0.6165
	Gain in quality of life	5%			Gain in quality of life	25%			Gain in quality of life	25%	
	Quality of life after	80%	0.0600		Quality of life after	80%	0.3650		Quality of life after	80%	0.8200
	Gain in life expectancy	0 years			Gain in life expectancy	5 years			Gain in life expectancy	10 years	
	Life expectancy after	15 years			Life expectancy after	30 years			Life expectancy after	15 years	
	Age of the patient group	10 years	-0.0375		Age of the patient group	55 years	-0.2954		Age of the patient group	55 years	-0.2954
	Alternative treatment	Yes, treats disease	-0.1112		Alternative treatment	Yes, treats disease	-0.1112		Alternative treatment	Yes, treats symptoms	-0.0284
	Total utility treatment		**0.1818**		Total utility		**0.5750**		Total utility		**1.1128**

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 6	Total costs	40M		Treatment 4	Total costs	20M		Treatment 8	Total costs	20M	
	Gain in quality of life	0%	0.1265		Gain in quality of life	5%	0.2705		Gain in quality of life	5%	0.2705
	Quality of life after	55%			Quality of life after	80%			Quality of life after	80%	
	Gain in life expectancy	0.5 years	0.0562		Gain in life expectancy	1 year	0.1162		Gain in life expectancy	5 years	0.4850
	Life expectancy after	1.5 years			Life expectancy after	6 years			Life expectancy after	6 years	
	Age of the patient group	55 years	-0.2954		Age of the patient group	35 years	-0.1628		Age of the patient group	75 years	-0.4568
	Alternative treatment	No	0		Alternative treatment	Yes, treats symptoms	-0.0284		Alternative treatment	No	0
	Total utility treatment		**-0.1127**		Total utility		**0.1956**		Total utility		**0.2988**

Portfolio 1				Portfolio 2				Portfolio 3			
Chosen treat-ments	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**	**Chosen treat-ments**	**Attributes**	**Attribute level**	**Utility**
Treatment 8	Total costs	20M		Treatment 7	Total costs	2M					
	Gain in quality of life	5%	0.2705		Gain in quality of life	5%	0.2705				
	Quality of life after	80%			Quality of life after	80%					
	Gain in life expectancy	5 years	0.4850		Gain in life expectancy	15 years	0.8550				
	Life expec-tancy after	6 years			Life expec-tancy after	30 years					
	Age of the patient group	75 years	-0.4568		Age of the patient group	35 years	-0.1628				
	Alternative treatment	No	0		Alternative treatment	Yes, treats symptoms	-0.0284				
	Total utility treatment		**0.2988**		Total utility		**0.9344**				
Additional savings (in addition to the 100M)		0M	0	Additional savings (in addition to the 100M)		3M	-0.0288	Additional savings (in addition to the 100M)		10M	-0.0960
Total utility lost by disinvesting portfolio			**0.5634**	**Total utility lost by disinvesting portfolio**			**2.9125**	**Total utility lost by disinvesting portfolio**			**2.6863**

7

General Discussion



Despite of the potential merits of disinvestment (i.e. fully or partially stopping the reimbursement of healthcare interventions) with regards to gains in health and containing healthcare expenditures, disinvestment decisions have been perceived to be challenging. Therefore, I hypothesized that specific mechanisms and considerations may be at play in disinvestment decisions, affecting the perceived difficulty and the feasibility of these decisions. The objectives of this thesis were 1) to obtain insight in the specific mechanisms and considerations that are relevant in disinvestment decisions, and 2) to investigate aspects affecting the feasibility of these decisions.

In the studies described in this thesis, I found that:

- The outcome of disinvestment processes is affected by the contextual factors support from the public and stakeholders (e.g. healthcare providers and policymakers), institutional role and financial interests of stakeholders, the organizational skills of patient groups, and the possibility to relieve the consequences of disinvestment for current patients (Chapter 2).

- The formal assessment criteria for the reimbursement of healthcare interventions are not consistently applied in disinvestment decisions (Chapter 2).

- There are four distinct societal viewpoints on disinvestment in the Netherlands that differ on their support for disinvestment and on the considerations that are found relevant in disinvestment decisions (Chapter 3). These four viewpoints are each supported by approximately half of Dutch citizens (Chapter 4).

- Common aspects considered relevant by those holding the different viewpoints are transparency of decision-making and medical necessity of the concerned healthcare intervention (Chapter 3).

- Resistance to disinvestment may partly be explained by the consequences of disinvestment citizens anticipate to experience themselves (i.e. the degree to which they will lose treatment options) as well as by not considering the increase in healthcare expenditure a large problem (Chapter 4).

- To compensate for the disinvestment of healthcare interventions, 1.58 to 1.86 times higher savings are required compared to the amount of money that people are willing to pay to reimburse these healthcare interventions in the first instance, indicating that loss aversion may play a role in disinvestment decisions (Chapter 5).

- Dutch citizens prefer to disinvest those healthcare interventions with limited to no health gain and that are targeted at patients of older age (Chapter 6).

In the remainder of this general discussion, I will discuss the role of the context in disinvestment decisions, the potential value of a consistently applied assessment framework, and why disinvestment decision-making remains a delicate task. Furthermore, I will discuss directions for further research and the potential impact that the Covid-19 pandemic may have on future disinvestment processes. Finally, I will end this discussion with the main conclusions of this thesis.

The context is key

The studies described in this thesis, consistently showed that the context is of key importance in disinvestment decisions. For instance, Chapter 2 indicates that contextual aspects, such as support among the public, policymakers and healthcare providers, are much more important for the outcome of disinvestment processes than the formal assessment criteria for the reimbursement of healthcare interventions. This has also been described in the literature in which a lack of support from stakeholders, a lack of the right financial incentives, and institutional factors have been described as important barriers to the implementation and success of disinvestment programmes [1-7].

One important aspect affecting the support of stakeholders for disinvestment, and as such, the outcome of the disinvestment process, is the view of stakeholders on the disease and healthcare intervention concerned [Chapter 2]. When the exact healthcare intervention and disease concerned is not known, stakeholders are very well able to determine the objective criteria they consider important in the disinvestment of healthcare interventions [Chapter 6]. However, in practice, the disease (i.e. patient group affected) and healthcare intervention concerned is always known. This may affect the outcome of disinvestment decisions to such a degree that the objective decision criteria may no longer be considered relevant [Chapter 2 and 6].

A phenomenon that may explain the importance attached to the disease and healthcare intervention concerned in disinvestment decisions is the identifiable victim effect. This effect refers to the observation that people are willing to take expensive measures to save identified victims, while they are willing to spend much less money on saving non-identified victims [8]. This effect is part of the rule of rescue, which is the imperative to rescue the life of identifiable individuals irrespective of its opportunity costs [9]. For this reason, the identifiable victim effect applies most strongly to situations where lives are at stake [8, 9]. Nevertheless, it also applies to situations in which only the quality of life of patients is affected [8, 9]. This identifiable victim effect has been observed in previous studies from the investment context (i.e. (not) starting reimbursement). These studies describe that media coverage made the patients affected by the potential denial of reimbursement of a healthcare intervention identifiable, resulting in the reimbursement of this healthcare intervention [10, 11].

With regards to disinvestment, the identifiable victim effect indicates that if patients affected by disinvestment are better identifiable, stakeholders become more reluctant to disinvest the healthcare intervention concerned, affecting the likelihood that disinvestment actually takes place. This is in line with the findings from previous research and the studies described in this thesis [Chapter 2, 3 and 6; 12, 13].

Some patient groups or treatments attract more sympathetic attention (i.e. positive attention leading to sympathy) than others [Chapter 2; 11]. For instance, cancer patients may attract much sympathetic attention, as compared to patients with mental health problems (e.g. addiction and schizophrenia) or diseases that are considered taboo (e.g. constipation). The degree to which a disease attracts sympathetic attention may affect the view stakeholders have on the patient group and treatment concerned, and, subsequently, the degree to which they identify with the patients [Chapter 2; 11]. As a result, patient groups that do not attract much sympathetic attention may be less identifiable to stakeholders with the consequence that they are more likely to have their treatment disinvested than patient groups attracting much sympathetic attention.

Another aspect affecting the identifiability of patient groups is their ability to make themselves visible. The ability of patient groups to exert pressure, i.e. to make themselves visible in the public debate, depends on their ability to organize themselves, which is affected by characteristics of the disease patients have [Chapter 2]. For instance, patient groups that are vulnerable as a result of their disease or the societal problems associated with their disease, may not be able organize themselves sufficiently enough to make themselves visible [Chapter 2]. This may also affect the likelihood of their case being covered in the media. As a result, these vulnerable patient groups may be less identifiable for stakeholders than less vulnerable patient groups, increasing the likelihood that their treatment is disinvested.

Summarizing, patient groups that are less vulnerable or that have a disease that attracts more positive attention are less likely to have their treatment disinvested than patient groups that are more vulnerable or have a disease that attracts less positive attention. From the principle of justice, we should not discriminate between people on morally irrelevant grounds [9]. Therefore, to determine whether differences in disinvestment on the grounds of vulnerability or having a sympathetic disease are justifiable, we should determine whether these grounds are morally relevant to discriminate on for the disinvestment of healthcare interventions. In my opinion, morally relevant grounds to discriminate on for the disinvestment of healthcare interventions should indicate the value the healthcare intervention has to patients and to society as a whole. I believe that the degree to which a treatment attracts positive (i.e. sympathetic) attention is not a valid ground to discriminate on with regards to disinvestment decisions. Vulnerability may be an indicator of the value of a healthcare intervention to patients and society, and, as such, a morally relevant ground to discriminate on with regards to disinvestment decisions. Prioritizing vulnerable patient groups may be justified to increase equity or to allocate the healthcare intervention to patients who need it most [14, 15]. However, when used to de-prioritize vulnerable patients, I believe that vulnerability is not a morally relevant ground to discriminate on for the disinvestment of healthcare interventions.

Therefore, to prevent unfair differences between patient groups with regards to the disinvestment of healthcare interventions, policymakers should try to limit the effect of contextual factors, such as the degree to which stakeholders identify with the patient groups concerned, on disinvestment decisions. One way to do this is by consistently applying the same criteria for every patient group. By strictly adhering to these criteria, contextual factors may be given less of a chance to affect the outcome of disinvestment processes, limiting inequality between patient groups.

A well-considered assessment framework is needed for disinvestment decisions

Previous research found that a structured, evidence-based process is a facilitator for the implementation of disinvestment decisions [4]. Furthermore, the need for a well-considered, consistently applied assessment framework has also been emphasized in the accountability for reasonableness framework (A4R, [16]). The A4R framework includes four conditions that must be met for organizations to be accepted as legitimate authorities to distribute healthcare budgets fairly: Publicity (i.e. transparency), Relevance (i.e. consistent and reasonable rationales), Revision and Appeals (i.e. opportunity to appeal to a decision), and Regulative (i.e. regulation to ensure the former conditions) [16]. Hence, increasing the consistency in the rationale underlying disinvestment decisions increases the legitimacy and fairness of these decisions. Therefore, I recommend policymakers to develop and consistently apply a well-considered assessment framework for disinvestment decisions.

What does this thesis tell us about how such an assessment framework should look like? Criteria that citizens consider important in disinvestment decisions are, among others, medical necessity, effectiveness (i.e. health gain), disease severity, and age of the patient group affected [Chapters 3 and 6]. Furthermore, one may consider using a higher cost-effectiveness threshold for disinvestment decisions compared to investment decisions [Chapter 5]. However, the importance attached to these criteria and the operationalisation of these criteria differs between groups in society [Chapter 3]. Therefore, selecting the criteria that should be included in an assessment framework may not be that straightforward. For instance, strictly evaluating the effectiveness of healthcare interventions was supported by approximately half of Dutch citizens, indicating that the other half of Dutch citizens may resist to this [Chapter 4].

In line with the A4R framework, I recommend policymakers to include criteria in the assessment framework for disinvestment decisions that "*fair-minded people can agree are relevant to pursuing appropriate care under necessary resource constraints*" [16]. With this I mean, to include criteria in the assessment framework for disinvestment decisions that people, who are fair-minded to develop such an assessment framework, can agree on as relevant in the context of these decisions. This does not mean that all groups in society have to agree on the operationalization of these criteria or on the weight these criteria receive; they only have to agree on the potential relevance of these criteria for disinvestment decisions. This thesis already provides some guidance on criteria people agree on as relevant. For instance, medical necessity may be a good criterion to include in the assessment framework for disinvestment decisions as all groups in society agree on its relevance, even though they prefer different ways to operationalize this criterion [Chapter 3]. To identify further criteria people can agree on as relevant with regards to disinvestment decisions, I recommend researchers to conduct citizen councils with citizens having different viewpoints on disinvestment to identify the criteria these citizens can agree on as relevant. The information obtained from such citizen councils can complement the information obtained in this thesis and can be of value as input for the development of a well-considered assessment framework for disinvestment decisions. A similar approach may also be applied to other stakeholder groups (e.g.

healthcare providers) to also obtain their input for the assessment framework. I will elaborate further on the process of developing such an assessment framework in the next section of this general discussion.

After the assessment framework has been developed, it is important to demonstrate to stakeholders that this framework is consistently applied, i.e. to make disinvestment decisions in a transparent way. Transparency is not only important to be able to demonstrate consistency; it has also been considered important in disinvestment decision-making by many citizens [Chapter 3 and 4]. Furthermore, it has been included in the A4R framework as a condition that must be met for legitimate and fair coverage decisions [16]. One main reason for this is that an advantage of transparent decision-making is that a decision-pattern may emerge [16]. In the long run, such a decision-pattern may increase the acceptability of disinvestment decisions for stakeholders because they can see that the case concerned is not treated differently from previous disinvestment cases. In the interviews conducted in the context of Chapter 2, several respondents mentioned the disinvestment process of the case concerned to be plotted as ‘a trial balloon’, just to see how the public would react to disinvestment. They felt that their case was the first that was appraised that critically, resulting in feelings of unfairness and anger. Hence, at the start, consistent and transparent disinvestment decision-making may still be difficult, with stakeholders considering their case a trial balloon. However, after a while, when a decision-pattern has emerged, disinvestment decisions may become more feasible as stakeholders may no longer feel they are treated unfairly, no longer considering their case to be a trial balloon.

Disinvestment remains a delicate task

Despite of the solutions proposed in the previous paragraphs to increase the feasibility of disinvestment decisions, making such decisions may still be considered difficult. A possible explanation for this can be found in the phenomenon of loss aversion. Loss aversion is the disutility people perceive from parting with a good in their endowment [17]. This effect has also shown to be present for goods that are included in the public endowment [18], which is also the case for the reimbursement of healthcare interventions. As disinvestment means that we have to part with a good in our public endowment, i.e. the reimbursement of a particular healthcare intervention, loss aversion may explain why disinvestment may make us feel unease, even when we know that the healthcare intervention has no effect or even is harmful [Chapter 5].

Another reason why disinvestment may still be considered difficult is that, despite of the transparent and consistent application of a well-considered assessment framework, some citizens may still oppose disinvestment. One reason for this is that there may always be a group in society that cannot be easily convinced of the necessity of disinvestment [Chapters 4 and 6]. These citizens may not consider the rising healthcare expenditures problematic or may feel that money could be easily saved elsewhere to finance the increase in healthcare expenditures [Chapter 4]. Also, they may not be aware of the fact that healthcare capacity (e.g. the labour force) is scarce and cannot be stretched endlessly to provide healthcare for every possible need. Furthermore, these citizens may feel indignant by the perceived deprivation of people's rights to healthcare as a result of disinvestment [Chapter 4 and 6]. To increase the support for disinvestment among these citizens, it is important to better explain to them why disinvestment is necessary.

A first approach to better explain to citizens why disinvestment is necessary is to make them aware of the opportunity costs of (not-)disinvesting healthcare interventions. With opportunity costs I mean the displacement taking place within the healthcare system [19]: using the scarce healthcare capacity to provide healthcare interventions with limited to no value (i.e. that should be disinvested), results in this capacity being no longer available for the provision of other types of healthcare that have more value. Hence, not disinvesting a healthcare intervention may result in the deprivation of healthcare elsewhere in the system where it may produce much more health. Making the unidentified patients affected by not-disinvesting healthcare interventions visible, may make citizens more aware of the necessity of disinvestment, showing them that it is not just an unpleasant task, but that it can help us to use the scarce healthcare capacity where it can be of most value.

Another approach to make citizens more aware of the need for disinvestment may be to explain to people that the reimbursement of healthcare interventions is directly related to healthcare expenditure and, subsequently, to the health insurance premiums they have to pay. Few people are aware that the annual increase in health insurance premiums is partly caused by the fact that the reimbursement package is extended with the reimbursement of new healthcare interventions. Therefore, I believe that it is important to explain to people that if more healthcare interventions are reimbursed, this will result in higher health insurance premiums. This may increase the perceived necessity to think critically on which healthcare interventions should and should not/ no longer be reimbursed.

Yet, another aspect complicating disinvestment decisions is that the transparent and consistent application of the to be developed, well-considered assessment framework by policymakers may not be of much value if the Minister of Health, who makes the final disinvestment decisions in the Netherlands, lets factors such as support among his/her voters and political pressure prevail over such an assessment framework. An example of this was seen for one of the cases in Chapter 2. In this case, pressure from healthcare providers, who the Minister needed for the implementation of other policies, was crucial for the decision that the healthcare intervention would not be disinvested. As described in the previous paragraphs, letting such contextual aspects prevail over a well-considered assessment framework, may lead to unfair differences between patient groups based on their vulnerability or the degree to which their disease attracts sympathetic attention. Therefore, I believe that it is important that the disinvestment process is de-politized. With this, I mean that I believe that disinvestment decisions should be made by policymakers that are further away from the political arena and, therefore, less susceptible to political pressure and other contextual factors than the Minister of Health currently is. Nevertheless, for the acceptability and legitimacy of disinvestment decisions, it may be important that a well-considered assessment framework for disinvestment decisions receives broad societal support, both among citizens and their political representatives. Therefore, I recommend policymakers to also involve politicians in the development of the assessment framework. In such a way, there is commitment of politicians to the assessment framework to be developed, making it easier to shift the execution of disinvestment decisions (i.e. making these decisions using the framework) to policymakers further away from the political arena.

Summarizing, disinvestment decision-making remains a delicate task, although there are approaches to increase its feasibility, such as making citizens more aware of displacement and shifting the responsibility for disinvestment from the Minister of Health to policymakers that are further away from the political arena. Nevertheless, disinvestment decision-making always remains a delicate task. Therefore, after having consulted the public and other stakeholders in the process of making disinvestment decisions, it is important that policymakers do not let loss aversion and resistance within society withhold them from actually making these decisions. In the end, it is the job of policymakers to make difficult decisions if these are considered to be in the best interest of society.

Directions for future research

In Chapter 2, we obtained insight in five cases of disinvestment using an interview study. These cases where selected from a longer list of 34 cases that were considered for disinvestment in the previous 10 years, enabling me to study a distinct set of cases in-depth. To obtain more insight in disinvestment processes beyond the five cases studied in this Chapter, I recommend future research to replicate the interview study to other cases and other settings. If researchers consider conducting a large number of interviews infeasible in their setting or for the cases selected, they may consider to conduct a qualitative content analysis of relevant documents as has been conducted by Embrett and Randall [20], instead of a more time-consuming interview study. However, as some relevant information is not written down in documents, assessing documents instead of conducting interviews may result in the omission of relevant information with regards to the disinvestment process of the selected cases.

In Chapter 3 to 6, we mainly focused on the preferences of citizens. Therefore, it may be interesting to also study the preferences of other stakeholder groups, such as patients and healthcare providers, and to assess whether their preferences differ from those of citizens. To this end, future research may consider conducting a Q-methodology study to assess the viewpoints of these other stakeholder groups on disinvestment. Furthermore, researchers may consider using preference elicitation techniques such as participatory value evaluation or discrete-choice experiments to measure the relative importance of a number of considerations in the context of disinvestment to these stakeholder groups. These studies may provide further insight in the resistance against disinvestment among these stakeholder groups and may provide input for the development of an assessment framework for disinvestment decisions.

Moreover, in Chapter 3 to 6, we elicited public views and preferences with regards to disinvestment, without referring to the exact disease and healthcare intervention concerned. As this thesis suggests that knowing the disease and healthcare intervention concerned may have a large impact on the preferences of citizens with regards to disinvestment, I recommend future research to assess the actual impact of such information on the preferences of citizens. To this end, a participatory value evaluation containing specific healthcare interventions for specific diseases as policy options may be conducted.

The potential impact of the Covid-19 pandemic on disinvestment

At the time the general discussion of this thesis was written (i.e. February and March 2021), the world is battling the Covid-19 pandemic. The world has been suffering from this pandemic for a year, affecting healthcare systems worldwide. In the Netherlands, the current healthcare capacity has been insufficient to treat both Covid-19 patients as well as regular (i.e. non-Covid-19) patients, causing delays in the provision of regular healthcare [21, 22]. These delays have caused significant health losses with recent estimates from the Netherlands suggesting that between 34,000 and 87,000 QALYs (quality-adjusted life years, i.e. life years in full quality of life) have been lost by delays in only 28% of the specialist hospital care during the first wave of the Covid-19 pandemic (i.e. March-August 2020) [22]. Although the impact of the Covid-19 pandemic on disinvestment has not been studied in this thesis, it is worthwhile to discuss the potential implications of this large healthcare crisis on disinvestment in the future.

The current healthcare crisis may have made people more aware of the scarcity in healthcare with regards to healthcare capacity. On the one hand, this may limit disinvestment as increasing the current healthcare capacity to be better prepared for the current crisis and subsequent crises may have more priority over cuts in healthcare to better allocate the limited healthcare capacity. On the other hand, the increased awareness of the scarcity in healthcare may have made people more aware that choices have to be made to allocate the scarce healthcare capacity such that it may be of most value. Therefore, the Covid-19 pandemic and its corresponding healthcare crisis may have made people more aware of the necessity of disinvestment because of the displacement taking place in the healthcare system. It may even have made policymakers aware that difficult choices with regards to the allocation of healthcare capacity should no longer be postponed, potentially facilitating disinvestment decision-making.

Furthermore, during the current healthcare crisis, many patients were confronted with the delay or cancellation of their healthcare. This may have made people more aware of the negative consequences of denying healthcare, which may contribute to the perception that denying patients a certain treatment is very harmful and should be avoided, limiting the feasibility of disinvestment. On the other hand, the delays in regular care may have demonstrated that some healthcare interventions may not be as necessary as thought before. In fact, the delays in healthcare provision may be seen as a natural experiment in which we could assess what the effect of delaying or not providing a large number of healthcare interventions is. This provides insight in which healthcare interventions actually deliver value and which ones not [23]. Researchers have argued to use the insights from this natural experiment to identify healthcare interventions that are of low value, and to, subsequently, reduce the provision of these healthcare interventions [24, 25]. However, to obtain such insights in the value of healthcare interventions, it is important to carefully monitor and evaluate the effects of delaying healthcare. Therefore, I recommend policymakers and researchers to start monitoring and evaluating the effects of delaying healthcare as soon as possible to take advantage of this natural experiment. If we wait too long to start monitoring, we may miss out on this opportunity to increase the effectiveness and efficiency of healthcare.

Conclusions

In this thesis, I have explored the specific mechanisms and considerations that are relevant in disinvestment decisions. Furthermore, I have investigated aspects affecting the feasibility of disinvestment decisions. To finalize the general discussion, the main conclusions of this thesis are:

- Contextual factors are most important in disinvestment decisions. This may, however, result in unfair differences between patient groups, based on their vulnerability or the degree to which their disease attracts sympathetic (i.e. positive) attention.

- The consistent and transparent application of a well-considered assessment framework for disinvestment decisions is important to increase the fairness and acceptability of disinvestment decisions.

- Although disinvestment decision-making remains a delicate task, there are approaches to further increase its feasibility, such as making citizens more aware of displacement and shifting the responsibility for disinvestment decisions from the Minister to policymakers that are further away from the political arena.

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Summary
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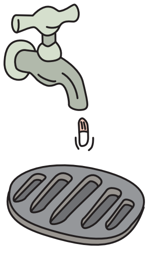

Summary

The persistent growth in healthcare expenditures has increased the pressure on the public financing of healthcare. To ensure that we can still afford our healthcare expenses in the future, this growth needs to be contained. One policy option that can contribute to this is the full or partial withdrawal of the reimbursement of healthcare interventions by means of a policy decision, also called disinvestment. Previous studies have estimated that 20 to 30% of the healthcare budget in OECD-countries[4] is spent on ineffective healthcare interventions (i.e. healthcare interventions that do not provide any gain in health or that are even harmful). For this reason, potentially, much can be gained from the disinvestment of healthcare interventions that are currently being used and reimbursed in the healthcare system. Despite of these potential merits, disinvestment decisions have been perceived as challenging. Even in cases where there was compelling evidence of ineffectiveness, proceeding to disinvestment has been shown to be very difficult. Specific mechanisms and considerations may, therefore, be at play, affecting the perceived difficulty and the feasibility of these decisions.

The objectives of this thesis were:

1. To obtain insight in the mechanisms and considerations that are relevant in disinvestment decisions.

2. To investigate aspects affecting the feasibility of disinvestment decisions.

Chapter 2 presents the findings of a qualitative interview study in which 37 semi-structured interviews were conducted with stakeholders (e.g. patients, policymakers, healthcare providers) who were involved in the policy process of five cases of disinvestment. This study aimed to obtain insight into the disinvestment processes for these cases, and to explore what aspects affected the outcome of these processes. The five distinct cases were selected from a list of 34 healthcare interventions that have been considered for disinvestment in the Netherlands in the previous 10 years. Some of the cases were eventually disinvested, while for other cases reimbursement was maintained. The interviews showed that support for disinvestment from stakeholders, especially from healthcare providers and policymakers, strongly affected the outcome of disinvestment processes. Furthermore, the institutional role of stakeholders as legitimized by the Dutch health insurance system, their financial interests in maintaining

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[4] OECD: The Organization of Economic Co-operation and Development

or discontinuing reimbursement, and the possibility to relieve the consequences of disinvestment for current patients affected the outcome of disinvestment processes as well. In the disinvested cases, patient groups were poorly organized, limiting their ability to exert pressure. No evidence was found of a consistent role of the formal Dutch package criteria (i.e. effectiveness, cost-effectiveness, necessity and feasibility) in disinvestment processes.

In Chapter 3, a Q-methodology study was conducted among a purposively selected sample of 43 citizens to explore the societal viewpoints in the Netherlands on the disinvestment of healthcare interventions. In this study, four viewpoints were identified. People holding viewpoint I believe that reimbursement of necessary healthcare should be maintained, irrespective of its costs. People holding viewpoint II agree with viewpoint I, although they believe that necessity should be objectively determined. People holding viewpoint III think that unnecessary, ineffective and inefficient healthcare should be disinvested. People holding viewpoint IV, consider it most important that disinvestment decision-making processes are transparent and consistent.

To obtain insight in the societal support for these four viewpoints, and to assess whether the support for these viewpoints is associated with background characteristics of citizens, we used an online survey among a representative sample of 1,794 adult citizens (Chapter 4). This survey showed that the support for each of the different viewpoints varied between 46.8% and 57.7% of the sample. Viewpoint support was associated with participants' age, gender, educational level, financial situation, healthcare utilization, opinion on the responsibility of the government for the health of citizens, and opinion on whether the increase in healthcare expenditures and health insurance premiums is considered a problem. From this, we concluded that resistance to disinvestment may partly be explained by the consequences of disinvestment citizens anticipate to experience themselves. Furthermore, citizens considering the increase in healthcare expenditure a larger problem are more supportive of disinvestment than those considering it less of a problem.

In Chapter 5, we reviewed the available evidence on the disparity between willingness-to-accept (WTA) and willingness-to-pay (WTP) for healthcare goods and services to obtain insight in whether losses in healthcare interventions are valued differently from gains in healthcare interventions. In this systematic review and meta-analysis, we included thirteen papers, reporting WTA and WTP from nineteen experiments/subgroups. The WTA/WTP ratios reported in these papers, varied from 0.60 to 4.01, with means of 1.73 (median 1.31) for fifteen estimates of the mean and 1.58 (median 1.00) for nine estimates of the median. Individual data obtained from six papers, covering 71.2% of the subjects included in the review, yielded an unadjusted WTA/WTP ratio of 1.86 (95% CI: 1.52-2.28) and a WTA/WTP ratio adjusted for age, gender and income of 1.70 (95% CI: 1.42-2.02). Subjects' income and age had a statistically significant effect on the WTA/WTP ratio. The approach to handling zero WTA and WTP values had a considerable impact on the WTA/WTP ratio found. The results of this Chapter imply that losses in healthcare interventions are valued differently from gains (ratio>1), indicating that loss aversion may play a role in disinvestment decisions.

To investigate the preferences of Dutch citizens towards the relative importance of attributes of healthcare interventions in the context of disinvestment in the Netherlands, a participatory value evaluation (PVE) was conducted in Chapter 6. In this PVE, a representative sample of 1,143 Dutch citizens was asked to select a number of healthcare interventions for disinvestment from a list of eight unlabelled healthcare interventions. These unlabelled healthcare interventions were described by the attributes age of the patient group, gain in quality of life, gain in life expectancy, availability of a treatment alternative, and costs/budget-impact. The participants were instructed to save at least €100 million. Results of the PVE showed that participants preferred to disinvest treatments resulting in smaller gains in quality of life and life expectancy (relative to larger gains) that are provided to older patient groups. Portfolio's (i.e. combinations of selected healthcare interventions) resulting in lower savings (i.e. the minimum required savings of €100 million) were preferred for disinvestment over portfolio's resulting in higher savings (i.e. >€100 million).

Finally, Chapter 7 discusses the main conclusions from the previous chapters, the overall conclusions of this thesis, directions for future research and the potential effect of the Covid-19 pandemic on future disinvestment processes. In this general discussion, I conclude that contextual factors are most important in disinvestment decisions. However, this large role of contextual factors may result in unfair differences between patient groups in the disinvestment of healthcare interventions, based on their vulnerability or the degree to which their disease attracts sympathetic attention (i.e. positive attention resulting in sympathy). Therefore, I argue that a well-considered assessment framework should be used in disinvestment decisions to limit the impact of contextual factors. The consistent and transparent application of such a well-considered assessment framework may increase the fairness and acceptability of disinvestment decisions. Nevertheless, disinvestment decision-making remains a delicate task. Fortunately, there are other approaches to increase the feasibility of disinvestment decisions, such as making citizens more aware of displacement resulting from not disinvesting a healthcare intervention (i.e. loss of healthcare elsewhere in the system to provide the non-disinvested healthcare intervention). Furthermore, we may consider to shift the responsibility for disinvestment from the Minister of Health to policymakers that are further away from the political arena, to limit the effect of contextual factors such as political pressure on the outcome of disinvestment decisions.

Nederlandse samenvatting

De aanhoudende groei in zorgkosten drukt in toenemende mate op de publieke financiering van de gezondheidszorg. Om ervoor te zorgen dat we onze zorgkosten in de toekomst nog steeds kunnen betalen, moet deze groei worden ingeperkt. Eén beleidsoptie die hieraan kan bijdragen is het gedeeltelijk of volledig stopzetten van de vergoeding van behandelingen door middel van een beleidsbeslissing, ook wel uitstroom genoemd. Eerdere onderzoeken hebben geschat dat 20 tot 30% van het zorgbudget in OESO-landen[5] besteed wordt aan ineffectieve behandelingen (i.e. behandelingen die geen enkele gezondheidswinst opleveren of die zelfs schadelijk zijn). Om deze reden kan er potentieel veel gewonnen worden met de uitstroom van behandelingen die op dit moment gebruikt en vergoed worden in de gezondheidszorg. Ondanks deze potentiele voordelen, worden besluiten om de vergoeding van zorg stop te zetten ervaren als zeer uitdagend. Zelfs in het geval van overtuigend bewijs van ineffectiviteit, is het erg lastig om over te gaan tot het stopzetten van vergoeding. Specifieke mechanismen en overwegingen spelen daarom waarschijnlijk een rol, wat waarschijnlijk invloed heeft op de ervaren moeilijkheid en haalbaarheid van deze besluiten.

De doelstellingen van dit proefschrift waren:

1. Inzicht verkrijgen in de mechanismen en overwegingen die relevant zijn bij besluiten om de vergoeding van behandelingen stop te zetten.

2. Te onderzoeken welke aspecten invloed hebben op de haalbaarheid van besluiten om de vergoeding van behandelingen stop te zetten.

Hoofdstuk 2 presenteert de resultaten van een kwalitatieve studie waarin 37 semigestructureerde interviews werden gevoerd met partijen (bijv. patiënten, beleidsmakers, zorgverleners) die betrokken waren bij het beleidsproces van vijf casussen van uitstroom. Het doel van deze studie was om inzicht te verkrijgen in de beleidsprocessen rond de uitstroom van deze vijf casussen. De verschillende casussen werden geselecteerd uit een lijst van 34 behandelingen waarvoor het stopzetten van vergoeding was overwogen in de laatste 10 jaar. Voor een aantal van de casussen werd de vergoeding uiteindelijk stopgezet, terwijl voor andere casussen de vergoeding uiteindelijk behouden bleef. De interviews lieten zien dat het draagvlak vanuit betrokkenen, met

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[5] OESO: Organisatie voor Economische Samenwerking en Ontwikkeling; in het Engels: Organisation of Economic Co-operation and Development (OECD)

name zorgverleners en beleidsmakers, een grote invloed had op de uitkomst van beleidsprocessen. Daarnaast, hadden ook de institutionele rol van betrokkenen zoals bepaald door het Nederlandse zorgverzekeringssysteem, de financiële belangen van betrokken met betrekking tot de vergoeding van zorg en de mogelijkheid om de gevolgen van uitstroom te verzachten voor huidige patiënten invloed op de uitkomst van het beleidsproces. In de casussen waarin de vergoeding werd stopgezet waren patiëntengroepen slecht georganiseerd, wat het vermogen van patiëntengroepen om druk uit te oefenen beperkte. Er werd geen bewijs gevonden voor een consistente rol van de Nederlandse pakketcriteria, zijnde effectiviteit, kosteneffectiviteit, noodzakelijkheid en uitvoerbaarheid, in de beleidsprocessen rondom het stopzetten van vergoeding van zorg.

In hoofdstuk 3 werd een Q-methodologie studie uitgevoerd onder een doelgericht geselecteerde steekproef van 43 burgers om te verkennen welke visies er in Nederland zijn over het stopzetten van de vergoeding van zorg. In dit onderzoek werden vier visies geïdentificeerd. Mensen die visie I ondersteunden, vonden dat de vergoeding van noodzakelijke behandelingen altijd vergoed moet blijven, onafhankelijk van de kosten. Mensen die visie II ondersteunden, waren het eens met visie I, alleen vonden zij wel dat noodzakelijkheid objectief moet worden aangetoond. Mensen die visie III ondersteunden, vonden dat de vergoeding van niet-noodzakelijke, niet-effectieve en niet-kosteneffectieve behandelingen stopgezet moet worden. Mensen die visie IV ondersteunden, vonden het vooral belangrijk dat besluiten over het stopzetten van vergoeding transparant en consistent genomen worden.

Om inzicht te verkrijgen in de maatschappelijk steun voor de vier visies over het stopzetten van vergoeding en om te bepalen of de steun voor deze vier visies samenhangt met de achtergrondkenmerken van burgers, hebben we in hoofdstuk 4 een online vragenlijst uitgezet onder een representatieve steekproef van 1794 volwassen burgers. Deze vragenlijst liet zien dat de steun voor elke visie varieerde tussen 46,8% en 57,7% van de steekproef. Steun voor de visies hing samen met de leeftijd, het geslacht, het opleidingsniveau, de financiële situatie en het zorggebruik van deelnemers. Ook hing het samen met de mening van deelnemers over de verantwoordelijkheid van de overheid voor de gezondheid van burgers en over de mate waarin de toenemende zorguitgaven en zorgpremies een probleem zijn. Hieruit concludeerden we dat weerstand tegen het stopzetten van vergoeding voor een deel kan verklaard worden door de gevolgen die burgers zelf verwachten te ervaren van het stopzetten van vergoeding. Daarnaast concludeerden we dat het stopzetten van vergoeding meer steun kreeg van burgers die de toename van zorgkosten een groter probleem vonden dan van burgers die het een kleiner probleem vonden.

In hoofdstuk 5 hebben we de beschikbare literatuur over het verschil tussen betalingsbereidheid (willingness-to-pay, WTP) en acceptatiebereidheid (willingness-to-accept, WTA) voor behandelingen in de zorg bekeken. Dit om na te gaan of verliezen in behandelingen anders worden gewaardeerd dan toenamen in behandelingen. In deze systematische review en meta-analyse hebben we dertien artikelen meegenomen die WTA en WTP rapporteren voor negentien experimenten/subgroepen. De WTA/WTP ratio's die in deze artikelen gerapporteerd werden, varieerden van 0,60 tot 4,01, met gemiddelden van 1,73 (mediaan: 1,31) voor vijftien schatters van de gemiddelde WTA/WTP ratio en van 1,58 (mediaan: 1,00) voor negen schatters van de mediane WTA/WTP ratio. Berekeningen aan individuele data afkomstig van studies beschreven in zes artikelen (omvat 71,2% van het totaal aantal mensen meegenomen in de review) resulteerden in een ongecorrigeerde WTA/WTP ratio van 1,86 (95% BI: 1,52-2,28) en een WTA/WTP ratio gecorrigeerd voor leeftijd, geslacht en inkomen van 1,70 (95% BI: 1,42-2,02). Het inkomen en de leeftijd van deelnemers had een statistisch significant effect op de WTA/WTP ratio. De gekozen aanpak om om te gaan met WTA en WTP waarden van nul had een aanzienlijk effect op de gevonden WTA/WTP ratio. De resultaten uit dit hoofdstuk impliceren dat verliezen in behandelingen anders worden gewaardeerd dan toenamen in behandelingen (ratio>1), wat erop wijst dat verlies aversie een rol kan spelen in besluiten over het stopzetten van vergoeding van behandelingen.

Om de voorkeuren van Nederlandse burgers met betrekking tot de relatieve belangrijkheid van attributen (i.e. kenmerken) van behandelingen in de context van uitstroom te onderzoeken, is er in hoofdstuk 6 een participatieve waarde-evaluatie (PWE) uitgevoerd. In deze PWE, is een representatieve steekproef van 1143 Nederlandse burgers gevraagd om een aantal behandelingen te selecteren uit een lijst van acht behandelingen die kandidaat waren voor uitstroom, om de vergoeding voor stop te zetten. Deze ongelabelde behandelingen werden beschreven met de attributen leeftijd van de patiëntengroep, winst in kwaliteit van leven, winst in levensverwachting, beschikbaarheid van een behandelalternatief en kosten/budgettaire impact. De deelnemers werden geïnstrueerd om tenminste €100 miljoen te besparen door het stopzetten van vergoeding. De resultaten van de PWE laten zien dat deelnemers een voorkeur hadden om de vergoeding stop te zetten van behandelingen met minder winst in kwaliteit van leven en levensverwachting (ten opzichte van meer winst) die gegeven worden aan oudere patiëntengroepen (ten opzichte van jongere patiëntengroepen).

Portfolio's (i.e. combinaties van geselecteerde behandelingen) die resulteerden in minder bezuinigingen (i.e. de minimaal gevraagde besparing van €100 miljoen) kregen de voorkeur voor uitstroom over portfolio's die resulteerden in meer bezuinigingen (i.e. >€100 miljoen).

Tenslotte bediscussieert hoofdstuk 7 de hoofdconclusies uit voorgaande hoofdstukken, de overkoepelende conclusies van dit proefschrift, suggesties voor toekomstig onderzoek en de potentiële impact van de Covid-19 pandemie op toekomstige beleidsprocessen rond het stopzetten van de vergoeding van zorg. In deze algemene discussie concludeer ik dat contextuele aspecten het meest belangrijk zijn in beleidsprocessen rond het stopzetten van de vergoeding van zorg. Deze belangrijke rol voor contextuele factoren resulteert echter in oneerlijke verschillen tussen patiëntengroepen met betrekking tot het stopzetten van de vergoeding van zorg, gebaseerd op de kwetsbaarheid van patiëntengroepen en de mate waarin hun ziekte sympathie oproept. Om de impact van contextuele factoren te beperken, pleit ik ervoor dat een weloverwogen afwegingskader gebruikt wordt in besluiten over het stopzetten van vergoeding. De consistente en transparante toepassing van zo'n weloverwogen afwegingskader leidt waarschijnlijk tot een eerlijker en beter geaccepteerd beleidsproces voor het stopzetten van vergoeding. Desondanks zullen besluiten om de vergoeding stop te zetten lastig blijven. Gelukkig zijn er andere benaderingen om uitstroom beslissingen haalbaarder te maken, zoals het meer bewust maken van burgers van verdringing in de zorg als gevolg van het niet stopzetten van vergoeding (i.e. een verlies van zorg elders in het zorgsysteem om de zorg waarvan de vergoeding niet is stopgezet, te kunnen blijven leveren). Daarnaast zouden we kunnen overwegen om de verantwoordelijkheid voor het stopzetten van de vergoeding te verplaatsen van de Minister van zorg naar beleidsmakers die verder weg zitten van de politieke arena, om zo de impact van contextuele factoren zoals politieke druk op de uitkomst van beslissingen over het stopzetten van vergoeding te beperken.

About the author

Adriënne Henderika Rotteveel was born on the 22nd of October 1991 in Haarlem. In 2010, she finished secondary school (Gymnasium degree) at the *Eerste Christelijk Lyceum* Haarlem, where she completed her economics exam one year earlier than the other students in her year. Adriënne then started the bachelor programme Health Sciences at the University of Twente, which she completed in 2013. Because of her interest in research in health services and health economics, Adriënne proceeded her studies at the University of Maastricht where she followed the master programme Health Services Research with the specialization Health Technology Assessment. During the second year of this master programme, Adriënne was an intern at the department of Clinical Epidemiology and Medical Technology Assessment (KEMTA) and the department of Surgery at Maastricht UMC+. During her internship, Adriënne focused on patient experiences with and treatment outcomes of the novel treatment option Sacral Neuromodulation for idiopathic slow-transit constipation. After obtaining her master degree with distinction in 2015, Adriënne continued working at the department of KEMTA as a junior researcher, where she contributed to the development of a medication-related patient reported experience measure (the PESaM, Patient Experience and Satisfaction with Medication, questionnaire) and the covenant phase of the conditional reimbursement program of the National Healthcare Institute for Sacral Neuromodulation for idiopathic slow-transit constipation. In 2016, Adriënne started a PhD at the National Institute for Public Health and the Environment (RIVM), UMC Utrecht and Erasmus University Rotterdam on disinvestment decisions in healthcare, which resulted in this dissertation. During her PhD, Adriënne has contributed to other RIVM projects as well, such as the project on the health effects of delays in regular healthcare resulting from the Covid-19 pandemic. After finishing her PhD dissertation, Adriënne continued working at the RIVM as a researcher in health economics and health services research.

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