HIGH-COST PATIENTS AND OPPORTUNITIES TO REDUCE UNNECESSARY SPENDING

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High-cost patients and opportunities to reduce unnecessary spending

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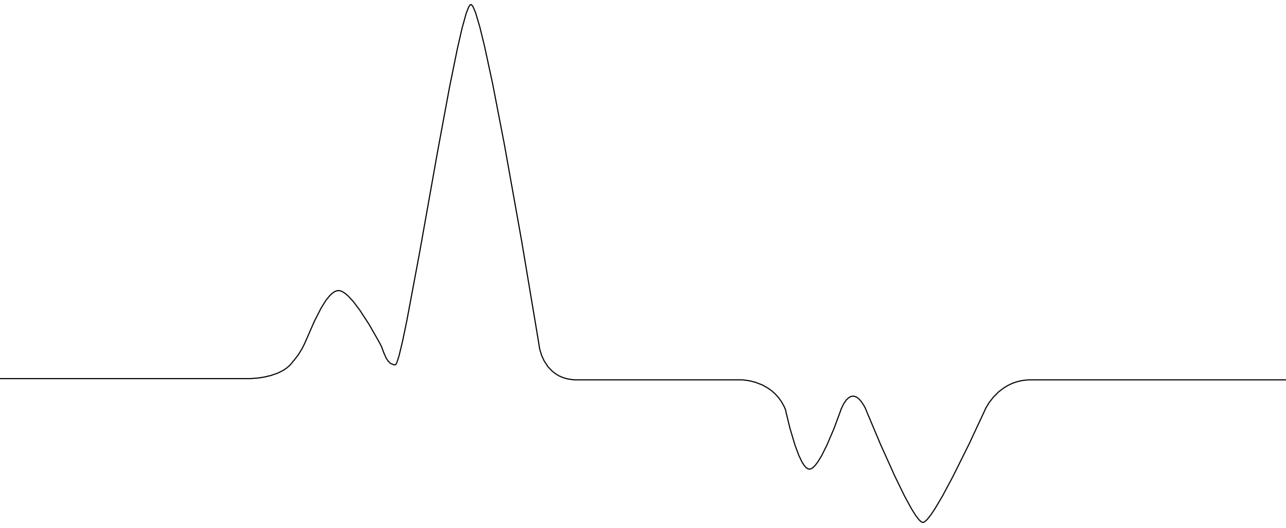
CHAPTER 1

General introduction and outline of the thesis



This thesis concerns the fiscal sustainability of healthcare. Although the continuing proliferation of our health system has brought tremendous benefit to our society, this has come at high costs, and the general consensus is that the forecasted growth in healthcare expenditures is unsustainable. There is thus ever more need for solutions to the cost problem of healthcare. The studies in this thesis explore two approaches for reducing unnecessary and harmful care while simultaneously improving outcomes of care. The first approach aims to track down unnecessary care and to identify the determinants of unnecessary care provision in Dutch healthcare settings; in order to effectively reduce unnecessary spending. The second approach encompasses an exploration of the characteristics and utilization of high-cost patients; the sickest patients who are in heaviest need for care, but who are most likely to receive suboptimal treatment and receive unnecessary care. Knowledge of this population is prerequisite for designing effective responses for increasing quality of care and reducing costs.

This introduction starts with a background in the problem of fiscal sustainability in the Netherlands, and a discussion of the Dutch approach to cost-containment. After this, the topic unnecessary care is introduced, including the two mentioned approaches for reducing unnecessary care. Subsequently, the goal and research questions, and a short overview of the contents of the thesis are presented.

Fiscal sustainability of healthcare

One of the most important achievements of modern Western countries has been the building of its comprehensive health systems. Patients receive high quality care for relatively low out-of-pocket costs at the point of care. As a result, people live longer and healthier lives than ever before. In the Netherlands, for example, life expectancy at birth has risen from 71.4 years in 1950 to 81.5 in 2016 [1]. In addition, life expectancy without physical limitations has risen from 65 years in 1983 to 72 years in 2012 for males, and from 64 years to 70 years for females [2]. Medicine has thus brought tremendous benefit to our society.

This has, however, come at high costs, and the growth of our health system has become a major fiscal burden. For as long as we know, it seems that healthcare costs can only grow, and grow, and grow. In the Netherlands, *collectively paid* healthcare costs have risen from 0.8% of gross domestic product (GDP) in 1950 to 9.3% of GDP in 2018 (figure 1). From the figure it can be inferred that from 2014 and onwards the increase in healthcare spending was outpaced by the growth in GDP.

According to a different indicator – used by Statistics Netherlands – *total* healthcare costs represented 13.8% of GDP in 2016 [3]. This means that at present, for families with modest incomes, healthcare costs account for about a quarter of their incomes. It has been projected that – with unchanged policy – in 2040 the same families with modest incomes may spend up to 40% of their incomes on healthcare [4]. In such scenario's, total healthcare costs grow up to €174 billion euro's in 2040 (€9.600 per capita spending, compared to €5.100 in 2015). The general consensus is that such an increase is unsustainable.

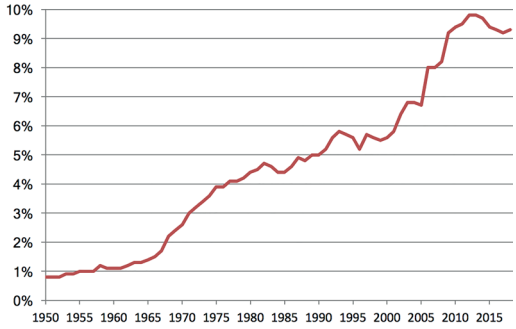


FIGURE 1 Collectively financed healthcare in the Netherlands as a percentage of GDP. Source: CPB, edited by Joost Wammes.

The financial crisis and healthcare spending

During the last decade, the global financial crisis has aggravated the problems of the fiscal sustainability of healthcare. In addition, due to the crisis the opportunity costs (the value of the choice of a best alternative cost while making a decision, in this case other public expenses; euro's can be spent only once) of increased spending on healthcare have increasingly become visible, putting ever more pressure on the health system.

Figure 2 shows the Dutch reaction to the global financial crisis. Dutch spending on healthcare and social security steadily rose as a proportion of GDP until 2013. The figure also shows that total public expenses as a share of GDP have continually dropped from 2010 and onwards; and much of this decrease in spending has been accounted for by budget cuts in public administration, defense and infrastructure. Honorary professor of Economics of the public sector Flip de Kam has referred to this when he called Dutch healthcare the 'cuckoo in the nest' ("koekoeksjong") of public spending [5]: budget cuts in other public sectors were needed to accommodate the growth of healthcare costs.

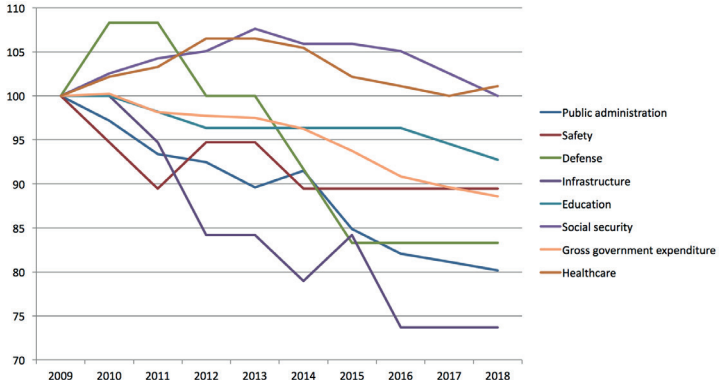


FIGURE 2 Public expenditures as a share of GDP (2009 proportion = 100). Source: CPB, edited by Joost Wammes.

Recent cost-containment measures in Dutch healthcare

In the Dutch curative health system, the main approach to controlling costs relies on market forces while regulating competition and improving efficiency of care (see chapter 2: Dutch healthcare system. NB: this thesis was limited to the curative health system, the long term care system was neglected). In addition, provider payment reforms, including a shift from a budget-oriented reimbursement system to a performance- and outcome-driven approach, have been implemented. In light of the global financial crisis, many additional activities have been taken to contain costs. In the following paragraphs the main ingredients of the Dutch approach are summarized. These efforts have effectively limited spending growth; Edith Schippers (2010-2017) was the first Minister of Health who finished a year without exceeding the predetermined budget.

Covenants are at the heart of Dutch cost-containment. In 2011, a first agreement ("bestuurlijk hoofdlijnenakkoord") was signed by a collaborate of the ministry of health, hospitals, and insurers. This agreement set a (voluntary) ceiling for the annual growth of spending on hospital care between 2012 and 2015. When overall costs exceed that limit, the government has the ability to control spending via generic budget cuts (via the Macro Management Tool "macrobeheersingsinstrument"). In the following years, similar agreements were signed for the mental health and primary care sector. These agreements included an extra 1 percent spending growth allowance for primary care practices in 2014 and 1.5 percent in 2015–2017, provided they demonstrate that their services are a substitute for hospital care. The current Ministry of Health has been negotiating with the parties to extend the covenants. The current agreements will expire at the end of 2022. The agreement for hospital care dictates a maximum volume growth of 0.8% in 2019, 0.6% in 2020, 0.3% in 2021, and 0.0% in 2022 (table 1).

TABLE 1 Permitted volume growth in covenants.

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022
Hospital care	2.5%	2.5%	1.5%	1%	1%	1%	1.4%	0.8%	0.6%	0.3%	0.0%
Mental health	-	2.5%	1.5%	1%	1%	1%	-	1.3%	1.1%	0.9%	0.7%
Primary care	-	3.0%	2.5%	2.5%	2.5%	2.5%	2.5%	2.5%	2.5%	3%	3%

One heavily debated cost-containment measure has been the increase in cost-sharing. The annual deductible, which accounts for the majority of patient cost-sharing, more than doubled between 2008 and 2018, from €170 to €385. There are some worries that this increase has led to greater numbers of people abstaining from or postponing needed medical care [6].

The 2012 coalition agreement noted that the benefit package would be 'stringently' managed (outdated treatments that do not (longer) meet the benefit package criteria would be excluded from public coverage. In addition, new treatments would be assessed more unambiguously, coherently and consistently on basis of the criteria), and that (relative) cost-effectiveness would get a legal status to inform coverage decisions [7]. This legal status has not been enforced until today, and the general consensus is that the management of

the benefit package has not, and will not yield in substantial cost savings. For example, van der Wees et al. found that countries worldwide show little variation in the scope of benefits covered, and that it is difficult for policy makers to completely remove services from the essential benefit package. Nevertheless, health technology assessment is gaining in importance and is used mainly for decisions concerning the benefit package and the appropriate use of medical devices.

The pharmaceutical sector has contributed significantly to the decrease in growth of spending. Average prices for prescription drugs have declined, with – as a result of stiff procurement – reimbursement caps for the lowest-price generic contributing to the decrease in average price. During the last few years however, many new and expensive drugs have entered the Dutch market, which further aggravated the cost pressure on the Dutch health system. The former Dutch health minister has formulated an ambitious policy proposal aiming in part to limit the pharmaceutical industry's power over drug pricing. During the Dutch European Union presidency, the topic was successfully put on the European Union agenda. In addition, many activities have been initiated to strengthen Dutch purchasing power to decrease the prize of expensive drugs. The management of the basic benefit package also contributes to this strategy. For example, in two technology appraisals in 2016, the Health Care Institute advised that Pertuzumab (Perjeta®) and Pembrolizumab (Keytruda®) (two expensive oncolytics) should not qualify for reimbursement, unless the cost-effectiveness would be improved and budget impact would be lowered through lower negotiated prices [8,9]. Based on this advice, the Minister negotiated lower prices with the manufacturers and decided that the drugs would qualify for reimbursement to the end of 2019. The effectiveness of these policies however, remains to be seen.

To conclude, many activities have been taken to reduce the growth of healthcare spending in the Netherlands. Based on the outcomes of these measures (the actual spending), it is fair to say the measures have been successful. Many questions remain however, about the sustainability of the measures. For example, further limitation of the basic benefit package or increase of the deductible are politically unattractive. In addition, new covenants will dictate a further reduction in the growth of services, putting again more pressure to the system. There is thus ever more need for alternative solutions to the cost problem of healthcare. Below, the topic unnecessary care is introduced, including the two mentioned approaches for reducing unnecessary care.

Stewardship

In 2012, the International Health Policy (IHP) survey of the Commonwealth Fund started to assess the overuse of health services. More than half (57%) of the Dutch general practitioners (GPs) believed – almost as much as their German colleagues – that Dutch patients receive (much) too much medical care (not just from them as a general practitioner, but from all care providers, including medical specialists). In 2015, that percentage fell to 46%, and the Netherlands was in the third place with only German (61%) and Swiss (51%) colleagues who scored higher. These were remarkable observations, as GPs are best positioned to overview and assess the value of care throughout the system, giving credibility to their assessments.

Besides, the gatekeeper system is designed to prevent unnecessary care. This begs for the questions: is the gatekeeping function still working as intended? And what did the general practitioners mean when they stated that Dutch patients receive 'too much' care? What is meant with the phrase 'unnecessary care' or 'low-value services'? And in what type of providers does this unnecessary care typically prevail? And among what patients?

What is meant with 'unnecessary care' or low-value services?

There is no lack of typologies to discern lower value from high values practices. For example, (cost-) ineffective care, inappropriately timed care, duplicate testing, medical errors, overtreatment, 'avoidable' hospitalizations and emergency department visits, or care that is not in line with the patients' preferences could be categorized as unnecessary care. Verkerk et al. recently published a typology of low-value care, in order to guide de-implementation. According to Verkerk, low-value services could be categorized to proven *ineffective care*, *inefficient care*, and *unwanted care*. Ineffective care is of low-value from a medical perspective, including proven (cost-)ineffective care for a particular subgroup or condition, or services which benefits do not weigh up to the harms according to scientific standards. Inefficient care is of low-value from a societal perspective. This care is in essence effective for the targeted condition, but becomes of low-value through inefficient provision or inappropriate high intensity or duration. Unwanted care is of low-value from a patients' perspective. This is in its essence effective care, but becomes low-value because it doesn't solve the individual patients' problem or does not fit the individual patients' preferences [10].

Traditionally, lower value or lower quality of care has been classified into misuse, overuse or underuse of health care services [11]. *Underuse* is the failure to provide a healthcare service when it would have produced a favourable outcome for the patient. Although fixing underuse is generally related to increased costs, in circumstances it may result in lower costs, for example in case of underuse of preventive drugs (one pervasive problem has been the underuse of beta-blockers after myocardial infarction: it is well-known that beta-blockers reduce mortality and morbidity, both important drivers of costs). *Misuse* occurs when an appropriate service has been selected but a preventable complication occurs – for example avoidable complications after surgery – and the patient does not receive the full potential benefit of the service.

Intuitively, the term overuse is closest to the term unnecessary care. *Overuse* occurs when a healthcare service is provided under circumstances in which its potential for harm exceeds the possible benefit. Antibiotic treatment for treating colds are a well-known example of overuse. One problem with overuse is that it is very hard to measure, as it requires a strict definition for the appropriateness of a service, based on evidence that considers the balance between benefits and harms for a population or individuals [12]. Nevertheless, many scholars have estimated the prevalence of overuse in healthcare may represent up to 10% and 30% of provided services [13-15].

But how to find these? What services are overused?

In practice, it has been proven very hard to identify services that could always be considered overuse or of lower value. One prominent problem in overuse is that interventions which are

high-value for a given subpopulation may be inappropriately applied to other populations [16]. As said, overuse requires a strict definition for the appropriateness of a service, based on evidence that considers the balance between benefits and harms for a population or individuals. Such clinical information is rarely identifiable from electronic health records or claim databases, let alone patient preferences. In addition, for the treatment of individual patients, the recommendations in clinical practice guidelines may not provide the clear cut answers as their wordings suggest they would give. This problem also persists in Verkerk's typology as it does not cover services of unknown effectiveness. The prevalence of such care may be enormous, NHS evidence once estimated that for 50% of services the effectiveness is unknown [17].

In summary, uncertainty exists about the exact prevalence of low-value services in the Netherlands. In daily practice it is difficult to discern low-value services from higher-value services. Being successful in this is prerequisite for developing effective policy solutions for reducing low-value care. Such insight might inform future policy concerning the benefit package, or to rationalize local delivery systems or care programs.

Nevertheless, the first approach geared towards services that are known to be of low-value only partly addresses the problem of unnecessary spending. Below we introduce an alternative approach, which encompasses an exploration of the characteristics and utilization of high-cost patients; the sickest patients who are in heaviest need for care. We will demonstrate that high-cost patients are at high risk to receive suboptimal treatment and receive unnecessary care, and that such a patient-centric approach may offer alternative opportunities for intervention.

High-cost patients

If unnecessary care or low-value care is not easily identifiable from electronic health records or claim databases, how else to find these? In what patients would we expect unnecessary care to be most persistent?

We hypothesized that most low-value spending may be concentrated among so-called high-cost patients. For long it is known that healthcare costs are heavily concentrated: the top 1% high-cost patients in the Netherlands account for about a quarter of healthcare costs, the top 5% of high-cost patients for about halve of total costs (in these calculations, costs were limited to the Health Insurance Act).

Our hypothesis was inspired by the theoretical work of Avedis Donabedian and colleagues [18]. Figure 3 (left panel) presents the hypothetical relationship between resource expenditures and expected health improvement, in case of an "ideal physician". The curve clearly shows that the marginal improvements in health sharply decline when resource expenditures are higher.

Figure 3 (right panel) shows the same curve, but now including a curve for the relationship between resource expenditures and expected health improvement, but for the "nonideal physician". Obviously, the total expected health improvement for the nonideal physician is lower than for the ideal physician, at any given resource input. Besides, and most importantly, the ideal physician will not use any resources anymore when the maximum health benefit is

reached. On the contrary, the nonideal physician continues to use resources, and from R_{IU} and onwards the curve actually takes a *downward turn*, implying health loss at increasing costs. According to Donabedian and his colleagues

> *We provide for the occurrence of harmful care at all levels of resource input, though we postulate that both the magnitude and the probability of harm are larger when resource inputs are excessive.*

Lower value services are thus most likely to persist in situations of high resource input. This work was developed to describe the relationship between the performance of 'all the physicians in the community as they care for all patients of a particular kind'. The paper thus primarily takes physicians as unit of analysis. It does not rule out however, that the described relationships could be interpreted within patients as well.

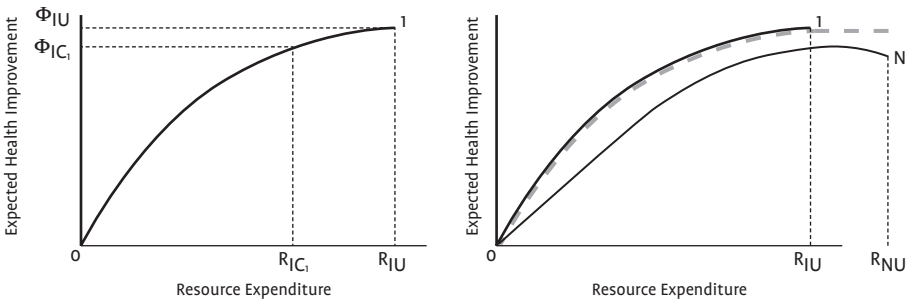


FIGURE 3 Left panel: Hypothetical relationship between resource expenditure and expected health improvement for strategies of care selected by the ideal physician. Right panel: Hypothetical relationships between resource expenditure and expected health improvement for strategies of care selected by the ideal physician and by the non-ideal physician. Source: Donabedian 1982.

There are other reasons why we would expect low-value services to concentrate among high-cost patients. By definition, high-cost patients receive most services and are thus most likely to experience problems with quality and safety in their care. Inherent to receiving many services are problems of coordination of care, or a general lack of integrated care delivery. In addition, medical care may be most complicated, and least supported with good evidence, among the sickest patients. It is widely known, for example, that clinical practice guidelines are written for 'artificial' patients with one single disease, while the majority of hospitalized patients suffer from several diseases. End of life periods are widely known for high costs, and Gilbert Welch, American medical doctor and author of the book 'Less medicine. More Health' once wrote [19].

> *How medical excess can be harmful is probably most familiar at the end of life. Here it is easy to see how aggressive intervention in the dying is not only futile, but inhumane. .... Medical excess is equally prevalent, however, at the other extreme of health: care for the well.*

In the US, high-cost patients have been increasingly studied, and many activities have been taken to improve care and reduce costs among high-cost patients. For example, Joynt et al. found that 10% of high-cost patients costs were deemed 'preventable' [20]. Policy and interventions aimed at high-cost patients, including for example care coordination and disease management, have had favourable results in quality of care and health outcomes, and mixed results in their ability to reduce hospital use and costs. Research has shown that the effectiveness and efficiency of the programs increase when interventions are targeted to the patients that most likely benefit. One such example is the 'hot spotting approach', named as such by Atul Gawande [21]

> *to look for the most expensive patients in the system and then direct resources and brainpower toward helping them*

In the Netherlands, high-cost patients have hardly been studied. In 2003, Polder estimated that the 1% costliest beneficiaries were responsible for 30% of spending in primary care, hospital care, pharmaceutical help and home and care. The costliest 5% and 10% were responsible for 55% and 70% of spending respectively. The costs in long term care were distributed even more unevenly [22]. A recent analysis by the CBS showed similar results: 20% of beneficiaries were responsible for 80% of health care spending [23]. Much uncertainty remains however, about the clinical characteristics and healthcare utilization patterns of high-cost patients in the Netherlands, but also in any other high-income countries. For example, although it is known that healthcare costs rise with increasing age, the proportion of non-elderly among high-cost populations is not studied. Besides, a major limitation of current literature is that little is known about patterns in care use and characteristics among different age groups, and this might offer clear resolution for policy making. In addition, to our knowledge, until today no studies have reported the role of expensive treatments (e.g. expensive drugs, transplant surgery, intensive care units, dialysis) as drivers of high costs. As shown above, the costs for expensive drugs have risen dramatically during the last few years. However, little is known about the relative contribution of expensive drugs towards the costs of high-cost patients. Much uncertainty persists also concerning the percentage of high-cost patients that are in their last year of life, and concerning the percentage of high-cost patients that persistently incur high costs (are patients incurring high costs in two or more consecutive years, or episodically). There is a lack of integral overview of drivers of high cost utilization and the relative importance of each driver among and across high-cost populations. Such information is prerequisite for developing tailored interventions aimed at high-cost patients, and to reconfigure the health system to best help the patients in heaviest need for high-quality care.

Rationale and goal of this thesis

Given the projections of future healthcare spending and that the opportunity costs of healthcare spending are increasingly visible, there is ever more need for alternative solutions to the cost problem of healthcare. Above we have shown that many types of low-value

services exist, that many scholars believe that the prevalence of lower value care may be high, so that reduction of such care may improve patient outcomes and reduce costs. The exact identification of such services however, has proven difficult, and we have shown that it is likely that most low-value care prevails among high-cost patients.
The research questions of this thesis are:

1 What are opportunities for cost-reduction through reduction of low-value services in the Netherlands?
2 What are the characteristics and healthcare utilization of high-cost patients and what strategies do likely improve high-cost patients care and reduce costs?

The goal of this thesis is to explore two approaches to cost containment. The first approach concerns the identification of agreed upon low-value services, or services that can be easily observed as having low value, and the context in which such care prevails. We conducted an exploratory survey among Dutch GPs to understand where the perceived unnecessary care prevails; and to identify factors that are associated with too much care at the entry point of Dutch healthcare. In addition, we developed an objective approach to identify and prioritize lower value services for practical de-adoption, and developed a list of lower value services identified from 193 Dutch clinical practice guidelines, published between 2010 and 2015. Furthermore, we interviewed 84 professionals to explore how Dutch health organizations have dealt with the cost pressure of cost-increasing and cost-ineffective health technologies, in order to inform future policy making concerning the introduction of new health technologies in the Dutch health system.

The second approach is very much related to the first. Here, we view the cost problem through a lens of high-cost patients, and study the characteristics and healthcare utilization of high-cost patients. Such knowledge is a first prerequisite for developing effective interventions and inform policy aimed at high-need, high-cost populations. We performed a systematic literature to synthesize the literature on high-cost patients' characteristics and healthcare utilization; determined the medical needs, demographic characteristics and healthcare utilization patterns of high-cost beneficiaries in the Netherlands; and studied the longitudinal healthcare utilization and characteristics of patients with heart failure and high costs.

Outline of this thesis

In this thesis we present six studies along our two specified approaches to cost reduction and healthcare improvement. **Chapter 2** gives an overview of the Dutch health system. Approach one – that aims to track down and reduce agreed-upon unnecessary care – includes chapter three until chapter five. **Chapter 3** presents an exploratory survey among Dutch GPs aiming to 1) understand where this perceived unnecessary care prevails; 2) identify factors that are associated with too much care at the formal entry point of Dutch healthcare. In doing this, we test assumptions supporting the gatekeeper system and further strengthening of this gatekeeper system. We asked the respondents to assess the

perceived amount of care by sector and type, and used several propositions describing factors possibly related to overuse and asked the respondents for opinion. **Chapter 4** describes the development of a list of lower value services identified from 193 Dutch clinical practice guidelines, published between 2010 and 2015. The list was developed with the aim to provide a comprehensive list of lower value services for Dutch hospital care. Furthermore, we compared our list with the original do-not-do – established by NICE in the UK – list on several aspects, including types of care and patient groups. Finally, we developed methods to prioritize the list on basis of several aspects, including prevalence of the disease and disease burden. **Chapter 5** presents an interview study after the introduction of cost-ineffective health technologies in the Netherlands. The aim of this research was to explore how Dutch health organizations have dealt with the cost pressure of cost-increasing and cost-ineffective health technologies. We conducted six case-studies and interviewed 84 professionals at all hierarchical levels (practitioners, departments, board of directors, insurers, and others) to explore the causality of resource allocation (how does one decision leads to another?) and the ultimate effects for individual patients.

Approach two – which encompasses an exploration of the characteristics and utilization of high-cost patients – includes chapter six until chapter eighth. **Chapter 6** presents our systematic review of high-cost patients' characteristics and healthcare utilization. We reviewed 55 studies of high-cost patients' characteristics and healthcare utilization. Andersen's behavioural model was used to categorize the characteristics of high-cost patients into predisposing, enabling and need characteristics. Our analysis was aimed at identifying drivers of costs that matter across payer types and countries. **Chapter 7** presents our Dutch claims database analysis on this issue. We first determined characteristics and spending and quantified the share of high-cost beneficiaries that use expensive treatments. We then used a beneficiary's most cost-incurring medical condition to examine characteristics and utilization patterns. In addition, we compared utilization and conditions across age groups. All analyses were performed for top-1% and top-2-5% beneficiaries separately. **Chapter 8** presents our second claims database analysis. In this study, we study the longitudinal healthcare utilization and characteristics of heart failure patients with high costs. We explore the characteristics of CHF patients with high costs and identified the determinants associated with high costs using generalized estimation equation modelling (GEE). Furthermore, we explore longitudinal healthcare utilization and determine the persistency of high costs within this population. **Chapter 9**, the discussion, summarizes the main findings of the thesis, discusses these findings in comparison with other research, and describes the implications of these findings for policy and practice.

References

1 Volksgezondheidenzorg.info. Trend in levensverwachting bij geboorte. Levensverwachting bij geboorte 1950-2016. 2018; Available from: https://www.volksgezondheidenzorg.info/onderwerp/levensverwachting/cijfers-context/trends#node-trend-levensverwachting-bij-geboorte

2 van Duin, C. and L. Stoeldraijer, Bevolkingstrends 2014. Projecties van de gezonde levensverwachting tot 2030. 2014, Centraal Bureau voor Statistiek.

3 CBS. Zorguitgaven stijgen in 2016 met 1,8 procent. 2017; Available from: https://www.cbs.nl/nl-nl/nieuws/2017/20/zorguitgaven-stijgen-in-2016-met-1-8-procent.

4 van der Horst, A., F. van Erp, and J. de Jong, Omgevingsscenario's voor gezondheid en zorg. 2011, CPB.

5 de Kam, F. Een koekoeksjong in Den Haag. 2010; Available from: https://www.nrc.nl/nieuws/2010/11/20/een-koekoeksjong-in-den-haag-11972303-a491485.

6 van Esch, T.E., et al., Increased cost sharing and changes in noncompliance with specialty referrals in The Netherlands. Health Policy, 2017. 121(2): p. 180-188.

7. Mastenbroek, C.G. and R.G.P. Doeschot, Stringent Pakketbeheer. 2012, CVZ.

8 Zorginstituut Nederland, Pakketadvies Pembrolizumab. 2016.

9 Zorginstituut Nederland, Pakketadvies Pertuzumab. 2016.

10 Verkerk, E.W., et al., Limit, lean or listen? A typology of low-value care that gives direction in de-implementation. Int J Qual Health Care, 2018.

11 Chassin, M.R. and R.W. Galvin, The urgent need to improve health care quality. Institute of Medicine National Roundtable on Health Care Quality. JAMA, 1998. 280(11): p. 1000-5.

12 Brownlee, S., et al., Evidence for overuse of medical services around the world. Lancet, 2017. 390(10090): p. 156-168.

13 Berwick, D.M. and A.D. Hackbarth, Eliminating waste in US health care. JAMA, 2012. 307(14): p. 1513-6.

14 Grol, R. and J. Grimshaw, From best evidence to best practice: effective implementation of change in patients' care. Lancet, 2003. 362(9391): p. 1225-30.

15 Morgan, D.J., et al., Setting a research agenda for medical overuse. BMJ, 2015. 351: p. h4534.

16 Scott, I.A. and A.G. Elshaug, Foregoing low-value care: how much evidence is needed to change beliefs? Intern Med J, 2013. 43(2): p. 107-9.

17 Garrow, J.S., What to do about CAM: How much of orthodox medicine is evidence based? BMJ, 2007. 335(7627): p. 951.

18 Donabedian, A., J.R. Wheeler, and L. Wyszewianski, Quality, cost, and health: an integrative model. Med Care, 1982. 20(10): p. 975-92.

19 Welch, G., Less medicine more health. 7 assumptions that drive too much medical care. 2015, Boston: Beacon press.

20 Joynt, K.E., et al., Contribution of preventable acute care spending to total spending for high-cost Medicare patients. JAMA, 2013. 309(24): p. 2572-8.

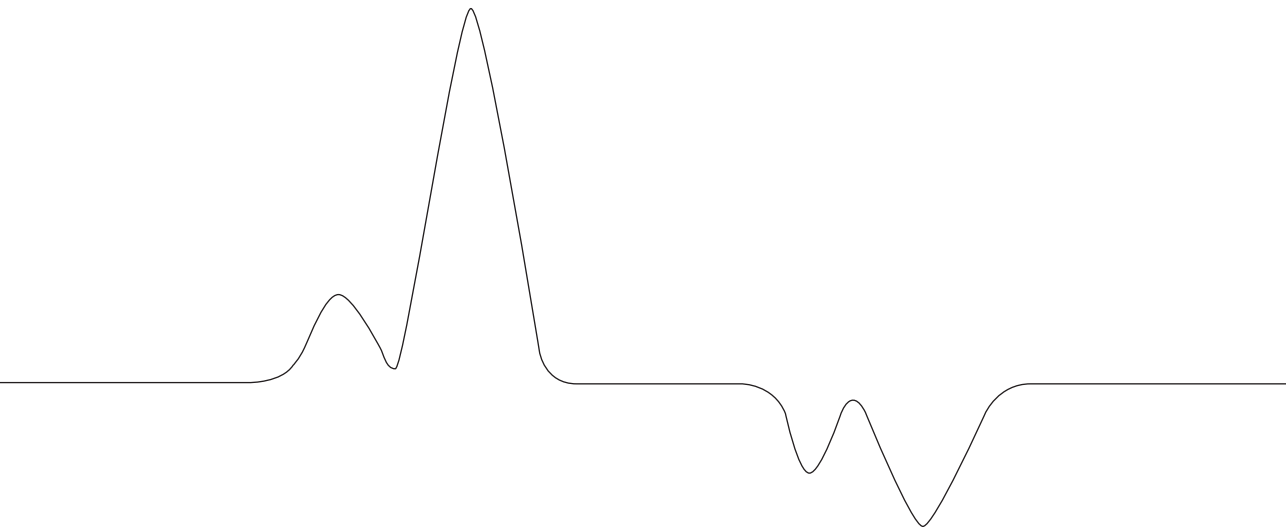
21 Gawande, A., The hot spotters: can we lower medical costs by giving the neediest patients better care?, in The New Yorker. 2011.

22 Polder, J., Het zorgstelsel is altijd solidair. ESB, 2003.

23 CBS, Gezondheid en zorg in cijfers. 2013.

CHAPTER 2

The Dutch Health Care System



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What is the role of government?

In the Netherlands, the national government has overall responsibility for setting health care priorities, introducing legislative changes when necessary, and monitoring access, quality, and costs. It also partly finances social health insurance (a comprehensive system with universal coverage) for the basic benefit package (through subsidies from general taxation and reallocation of payroll levies among insurers via a risk adjustment system) and the compulsory statutory health insurance system for long-term care. Prevention and social supports are not part of statutory health insurance but are financed through general taxation. Municipalities and health insurers are responsible for most outpatient long-term services, including personal and home care, and all youth care under a provision-based approach (with a high level of freedom at the local level).

Universal coverage

In the Netherlands, health insurance was installed in 1941 according to the German Bismarck model of public and private health insurers. Around 63% of the population was covered by public health insurance, while more affluent could opt for private insurance or choose to remain uninsured. At the turn of the century, concerns of inefficiencies and long waiting lists provided momentum for a market oriented reform inspired by the Enthoven proposal of managed competition. The 2006 Health Insurance Act (reform) merged the traditional public and private insurance into one universal social health insurance with mandatory coverage. In 2011, the government started to track down the uninsured. Since then, the number of uninsured has steadily declined, and by the end of 2016, 23,000 people (less than 0.2% of the population) remain uninsured.

How is the health system financed and who is covered?

Publicly financed health insurance In 2016, the Netherlands spent 10.5 percent of GDP on health care, and 81 percent was collectively financed, consisting of a mixture of insurance premiums (21%), copayments (11%), earmarked payroll taxes (46%) and general taxation (22%) [1]. All residents (and nonresidents who pay Dutch income tax) are mandated to purchase statutory health insurance from private insurers. Uninsured are fined and premiums may be levied directly from income. People who conscientiously object to insurance can opt out by making mandatory contributions into a health savings account. Active members of the armed forces (who are covered by the ministry of defense), are exempt. Insurers are required to accept all applicants, and enrolees have the right to change their insurer each year.

Apart from acute care, long-term care, and obstetric care, undocumented immigrants have to pay for most health care themselves (they cannot take out health insurance). However, some mechanisms are in place to reimburse costs that undocumented immigrants are unable to pay. For political asylum seekers, a separate set of policies has been developed. Permanent residents (for more than three months) are obliged to purchase private insurance

coverage. Visitors are required to purchase insurance for the duration of their visit if they are not covered through their home country.

Statutory health insurance is financed according to the 2006 Health Insurance Act, through a nationally defined, income-related contribution (6.9 percent of up to EUR 54,614 [USD 67,500] of annual taxable income) (45%), a government grant for children and youth aged 18 and under (5%), and community-rated premiums set by each insurer (45%). Children under 18 are automatically covered, while adults choose a policy on an individual basis (no family coverage). Adults with the same insurer pay the same premium, regardless of their age or health status. However, through employer collectives, lower premiums may be offered. Income taxes and government grants are collected in a central health insurance fund, and redistributed among insurers in accordance with a risk-adjusted capitation formula that considers age, gender, labor force status, region, and health risk (mostly based on past drug and hospital utilization).

Private, statutory insurers are expected to engage in strategic purchasing, and contracted providers are expected to compete on both quality and cost. There are 10 statutory insurers in 2018, but the insurance market is dominated by the four largest insurance conglomerates, which account for 90 percent of all enrollees. Currently, all insurers are mandated to operate as non-profits.

Private (voluntary) health insurance In addition to statutory coverage, most of the population (84%) purchases supplementary voluntary insurance covering a mixture of benefits not covered by statutory insurance, such as dental care, alternative medicine, physiotherapy, eyeglasses and lenses, contraceptives, as well as reduced copayments for on-label medicines (excess costs above the limit for equivalent drugs – an incentive for using generics). Premiums for voluntary insurance are not regulated; insurers are allowed to screen applicants based on risk factors. Nearly all of the insured purchase their voluntary benefits from the same (mostly nonprofit) insurer that provides their statutory health insurance. People with voluntary coverage do not receive faster access to any type of care, nor do they have increased choice of specialists or hospitals. In 2016, voluntary insurance accounted for 7.0 percent of total health spending.

Safety net GP care and children's health care up to the age of 18 are exempt from cost-sharing. Government also provides subsidies (health care allowances), subject to asset testing and income ceilings, to cover community-rated premiums for low-income families: singles with annual income less than EUR28,500 (USD35,200) and households with income less than EUR35,500 (USD44,000). Over 5 million people, or about 30 percent of the total population, receive allowances set on a sliding scale, ranging from EUR4.00 (USD5) to EUR94.00 (USD116) per month for singles and from EUR7.00 (USD9) to EUR 176.00 (USD 217) for households, depending on income.

What is covered?

Benefit package The government determines the statutory benefits package, guided by advice from the National Health Care Institute. The mandatory benefit package includes,

among other things, care provided by general practitioners (GPs), hospitals, and specialists; maternal care; dental care (up to age 18); prescription drugs; physiotherapy up to age 18; home nursing care; basic ambulatory mental health care for mild-to-moderate mental disorders; and specialized outpatient and inpatient mental care for complicated and severe mental disorders. Health insurers are legally required to provide these standard benefits. Since 2015, nursing home care for elderly and disabled is financed under the Long-Term Care Act (see below).

Some treatments, such as general physiotherapy and pelvic physiotherapy for urinary incontinence, are only partially covered for some people with specific chronic conditions, as are the first three attempts at in vitro fertilization. Some elective procedures, such as cosmetic plastic surgery without medical indication, dental care above age 18, and optometry and other vision care without medical indication, are excluded. A limited number of health promotion programs are covered, including smoking cessation and some weight management advice. A range of medical devices is covered, including hearing aids and orthopedic shoes, but wheelchairs and other walking aids are excluded. The Public Health Act describes municipal responsibilities for national prevention programs, vaccinations and infectious disease management. Municipalities can install additional prevention programs, such as healthy living and obesity reduction programs, but this varies wildly from one municipality to another.

As of 2015, home care is a shared responsibility of the national government, municipalities (day care, household services), and insurers (nursing care at home) and is financed through the Health Insurance Act and the Social Support Act (Wmo). Hospice care is financed through the Long-Term Care Act of 2015.

Cost-sharing and out-of-pocket spending For the Health insurance Act, the main form of cost sharing is a mandatory deductible of EUR385 (USD440) as of 2018. Children under 18 are exempt. In addition, consumers may take on a voluntary deductible of EUR500 (USD570). The deductible covers a broad range of health services, including hospital admissions, specialist services and prescription drugs. Some services are exempt, such as GP visits and preventative services, including most immunizations and breast cancer screening. For some selected services, such as on-label medications, physiotherapy, medical transportation or medical devices, additional cost sharing may be required via copayments, coinsurance, or direct payments. Patients with an in-kind insurance policy may be required to share costs of care from a provider that is not contracted by the insurance company. For long-term care, an income- and wealth-related copayment up to a maximum of EUR2,332 (USD 2,664) per month is required. For municipal home care and social services, most municipalities require a small income-related copayment. Out-of-pocket expenses represented 12.2 percent of health care spending in 2016.

How is the delivery system organized and how are providers financed?

Physicians: medical education and workforce The number of medical doctors is regulated through caps on the number of medical students, both at a national level and at a university

level. Medical schools are located in private, not-for-profit university medical centers. Regular medical students pay a yearly tuition fee of approximately €2,100. The Capacity Body (Capaciteitsorgaan) advises the Ministry of Health on all specialized postgraduate training programs for medical specialists to assure matching supply and demand. No such limitations apply for college level nursing educations. Regional bodies supervise nursing training programs, subsidized by the Ministry of Health. There are no national initiatives to ensure the supply of medical providers in rural or remote areas. However, in rural areas GPs may take over the role of pharmacists for the supply of prescription-only pharmaceuticals [2].

Primary care There were 13,364 registered primary care doctors (GPs) and 32,605 medical specialists in 2017. Forty-two percent of practicing GPs worked in group practices of three to seven, 40 percent worked in two-person practices, and 18 percent worked solo (2016). Most GPs work independently or in a self-employed partnership; one third are employed in a practice owned by another GP or are contracted on the basis of short-term contracts.

The GP is the central figure in the Dutch primary care. The typical practice size is approximately 2200 patients per full-time working GPs. Although registration with a GP is not formally required, most citizens (over 95%) are registered with one they have chosen, and patients can switch GPs as often as they like. GPs have a gatekeeping function; referrals are required for both hospital and specialist care.

Many GP practices employ salaried nurses and primary care psychologists. Primary care psychologists constitute specially trained psychologists, nurses and social-caregivers. Reimbursement for primary nursing care is received by the GP, so any productivity gains that result from substituting a nurse for a doctor accrue to the GP. Chronic care management is coordinated through care groups (mostly GP networks). Care groups are legal entities that assume clinical and financial responsibility for the chronic disease patients who are enrolled; the groups purchase services from multiple providers. To incentivize care coordination, bundled payments are provided for certain chronic diseases, such as diabetes, cardiovascular conditions, and chronic obstructive pulmonary disease (COPD).

In 2015, the government introduced a new GP funding model comprising three segments. Segment 1 (representing ≈75% of spending) funds core primary care services and consists of a capitation fee per registered patient, a consultation fee for GPs (including phone consultation), and consultation fees for ambulatory mental health care at the GP practice. The Dutch Health Care Authority (Nederlandse Zorgautoriteit) determines national provider fees for this segment. Segment 2 (≈15% of spending) consists of funding for programmatic multidisciplinary care for diabetes, asthma, and COPD, as well as for cardiovascular risk management; prices are negotiated with insurers. Segment 3 (≈10% of spending) provides GPs and insurers with the opportunity to negotiate additional contracts – including prices and volumes – for pay-for-performance and innovation. Primary care providers are not allowed to bill patients extra and above the fee schedule.

In 2018, self-employed GPs earn average gross annual income of EUR 135,000 (USD 167,000) (excluding out-hours care). In 2016, the gross annual incomes of specialists were estimated at maximally EUR 160,000 for salaried specialists (USD 197,000; ratio to GPs 1.2:1) and EUR 211.000 (USD 260,000; ratio to GPs 1.6:1) for independent specialists.

Outpatient specialist care Nearly all specialists are hospital-based and either part of group practices (39%) or on salary (49%, mostly in university clinics; the remaining 12% work both on salary and independently). Before 2015, a fixed part of hospital payments was reserved for medical specialists. As of 2015, specialist fees are freely negotiable between specialists and hospitals. This so-called "integral funding" dramatically changed the relationship between medical specialists and hospitals. Hospitals now have to negotiate in allocating their financial resources among their specialists. After patients receive referral for specialist treatment (in any hospital), patients are free to choose their provider, but insurers may set different conditions (e.g., cost-sharing) for different choices within their policies [3]. There is a nascent trend toward working outside of hospitals – for example, in growing numbers of (mostly multidisciplinary) ambulatory centers – but this shift is marginal, and most ambulatory centers remain tied to hospitals. Specialists in ambulatory centers tend to work most of the time in academic or general hospitals.

Administrative mechanisms for paying primary care doctors and specialists The annual deductible (which has to be paid for amongst other specialist physician visits, but not for GP visits, see above) is paid to the insurer. The insured have the option of paying the deductible before or after receiving health care and may choose to pay all at once or in installments. Other copayments – those for drugs or transportation, for example – have to be paid directly to the provider.

After-hours care After-hours care is organized at the municipal level in "GP posts," which are walk-in centers, typically run by a nearby hospital, that provide primary care between 5 p.m. and 8 a.m. Nearly all GPs work for a GP post. Specially trained doctor assistants answer the phone and perform triage; GPs decide whether patients need to be referred to a hospital. Doctors are separately compensated at hourly rates for after-hours care (on top of the regular income). At least 50 hours of after-hours care must be provided annually to maintain their registration as general practitioner. The GP post sends the information regarding a patient's visit to the patient's regular GP. Since out-of-hours care is typically provided at hospitals, there is no national medical telephone hotline advising patients on their nearest out-of-hours locations.

Hospitals In 2016, there were 79 hospital organizations, including eight university medical centers. All hospitals are private entities but profits may not be distributed to shareholders, making the hospital market virtually 100% private non-profit. In 2015, there were 231 independent private and nonprofit treatment centers whose services were limited to same-day admissions for non-acute, elective outpatient care (e.g., eye clinics, orthopedic surgery centers) covered by statutory insurance.

Hospital payment rates, through which doctors are paid, are mostly determined through negotiations between each insurer and each hospital over price, quality, and volume. The great majority of payments take place through the case-based diagnosis treatment combination system (DRG-like) called DBC defined by the Dutch Health Care Authority. The rates for approximately 70 percent of DBCs are freely negotiable (DBC-B segment); the remaining 30 percent are set nationally by the Dutch Health Care Authority (DBC-A segment).

The number of DBCs was reduced from 30,000 to 4,400 in 2012 to reduce administrative complexity. Since 2015, independent medical specialist groups negotiate with their hospital for their remuneration. Diagnosis treatment combinations cover both outpatient and inpatient as well as specialist costs, strengthening the integration of specialist care within the hospital organization.

A small part of hospital care is reimbursed through so-called add-ons. Add-ons are separate payments that have been developed for the reimbursement of expensive drugs and intensive care unit admissions. University medical centers receive special allowances (so-called 'academic component') for the adoption of new technologies.

Mental health care Mental health care is provided by specially trained psychologists, nurses and social caregivers in basic ambulatory care settings, such as GP offices, for mild-to-moderate mental disorders. In cases of complicated and severe mental disorders, GPs will often refer patients to basic mental health care (e.g., a psychologist or an independent psychotherapist) or to a specialized mental health care institution. Mental care delivered by GP offices or referred by GPs is generally covered as part of the basic benefit package. Inpatient mental care is covered as part of the Long-term Care Act. Hospitals provide acute mental care. The delivery of preventive mental health care is the responsibility of municipalities and is governed by the Social Support Act of 2015. For several years, policymakers have been aiming to substitute specialized and basic mental health care for primary mental health care, as reflected in the steady increase in the number of GPs who employ primary care psychologists.

Long-term care and social support Long-term care and social support operate as separate programs, complementary to the curative health system. Long-term care is financed through the Long-Term Care Act of 2015 (Wet langdurige zorg), a statutory social insurance scheme for long-term care and uninsurable medical risks and cost that cannot be reasonably borne by individuals. It operates nationally, and taxpayers pay a contribution of 9.65% of taxable income up to €33.791 in 2017. In 2017, a total of €20.0 billion was spent on long-term care under the Long-term Care Act. Home care services, youth care, ancillary services and social support services are financed by municipalities through the Social Support Act of 2015 (Wet maatschappelijke ondersteuning), about 6.5 billion in 2017. Municipalities receive block grants from the government, covering all municipal expenses. Municipalities have very limited tax-raising abilities.

Long-term care encompasses residential care; personal care, supervision, and nursing; medical aids; medical treatment; and transport services. Patients in need of permanent supervision, or patients who need assistance 24 hours per day to prevent escalation or serious harm, are eligible. The Centre for Needs Assessment (Centrum Indicatiestelling Zorg, CIZ), a regulatory governmental agency, determines eligibility based on clinical need alone (no means-testing). Cost sharing depends on annual income and wealth, age and household size. In 2017, 314,220 people used long-term care.

The Social Support Act provides funding through a block grant from the national government to municipalities, that are responsible for ensuring the provision of household services, medical aids, home modifications, services for informal caregivers, preventive

mental health care, transport facilities, and other assistance. Municipalities have a great deal of freedom in how they organize these services, including needs assessments, and in how they support caregivers (e.g., through the provision of respite care or a small allowance). In 2017, 1,042,790 people used Social Support services funded by municipalities.

Long-term care is provided by private, nonprofit organizations. For home care (since 2015 part of Health Insurance Act and Social Support Act) profits are allowed. Most palliative care, including hospice care, is integrated into the health system and can be delivered by general practitioners, home care providers, nursing homes, specialists, and volunteer workers. Palliative care is financed through a number of sources, but mostly through the Long-Term Care Act.

Under the Health Insurance Act of 2006, the Social Support Act of 2015 and the Long-Term Care Act of 2015, personal budgets are provided for patients to buy and organize their own (long-term) care. Under the Health Insurance Act and the Social Support Act, health insurers and municipalities are free to set "sufficient" budget rates (typically about 70% of in-kind rates), whereas under the Long-Term Care Act, budget rates are set nationally. Municipalities have a great deal of freedom in how to support family and informal caregivers, for example, through respite care or a small allowance.

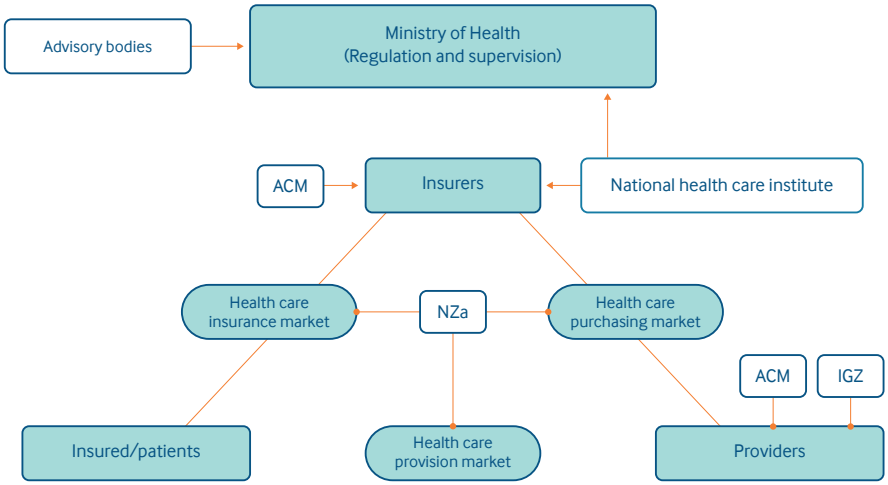


FIGURE 1 Organization of the Health System in the Netherlands. *Source: J. Wammes, P. Jeurissen, and G. Westert, Radboud University Medical Center, 2014.*

What are the key entities for health system governance?

Since 2006, the Ministry of Health's role has been to safeguard health care from a distance rather than managing it directly. It is responsible for the preconditions pertaining to access, quality, and cost in the health system, has overall responsibility for setting priorities, and may, when necessary, introduce legislation to set strategic priorities.

A number of arm's-length (independent) agencies are responsible for setting operational priorities. At the national level, the Health Council advises government on evidence-based medicine, health care, public health, and environmental protection. The National Health Care Institute advises the government on the components of the statutory benefits package and has various tasks relating to quality of care, professions and training, and the insurance system (e.g. risk adjustment). The Medicines Evaluation Board oversees the efficacy, safety, and quality of medicines. The National Health Care Institute assesses new technologies on effectiveness and cost effectiveness, and advises the Minister on uptake into the mandatory benefit package. Decisions about the benefits package rest with the health minister. The Dutch Health Care Authority (Nederlandse Zorgautoriteit) has primary responsibility for ensuring that the health insurance, health care purchasing, and care delivery markets all function appropriately – for example, by designing and managing the diagnosis treatment combination system and setting prices for 30 percent of diagnosis treatment combinations. Meanwhile, the Dutch Competition Authority (Autoriteit Consument en Markt) enforces antitrust laws among both insurers and providers. The Health Care Inspectorate supervises the quality, safety, and accessibility of care. Self-regulation by medical doctors is also an important aspect of the Dutch system [4]. Private insurers are tasked with increasing health system efficiency and cost control through prudent purchasing of health services.

Public engagement and public information are not centralized in one body. The patient rights movement consists of a wide range of organizations, some for specific diseases and some functioning as umbrella organizations. The patient umbrella organization (Patiëntenfederatie Nederland) conducts a range of activities to promote transparency.

Health information technology is not centralized in one body. The Union of Providers for Health Care Communication (De Vereniging van Zorgaanbieders voor Zorgcommunicatie) is responsible for the exchange of data via an information technology (IT) infrastructure.

Health equity has not been considered a policy priority by any organization or agency in the Netherlands.

What are the major strategies to ensure quality of care?

Private, statutory insurers are expected to engage in strategic purchasing, and contracted providers are expected to compete on both quality and cost. At the system level, quality is ensured through legislation governing professional performance, quality in health care institutions, patient rights, and health technologies. The Dutch Health Care Inspectorate is responsible for monitoring quality and safety. In 2014, the National Health Care Institute was established to further accelerate the process of quality improvement and evidence-based practice. As part of the National Health Care Institute, the National Quality Institute promotes quality measurement and transparency. Most quality assurance is carried out by providers, sometimes in close cooperation with patient and consumer organizations and insurers. There are ongoing experiments with disease management and integrated care programs for the chronically ill.

In the past few years, many parties have been working on quality registries. Most prominent among these are several cancer registries and surgical and orthopedic (implant)

registries. Mechanisms to ensure the quality of care provided by individual professionals include registration into a government-based national register, including re-registration every 5 years, contingent upon compulsory continuous medical education (the content is determined by professional organizations); regular on-site peer assessments by professional bodies; and professional clinical guidelines, indicators, and peer review. The main methods used to ensure quality in hospitals, nursing homes and other healthcare institutions include voluntary accreditation and certification granted by independent organizations); compulsory and voluntary performance assessment based on indicators; and national quality improvement programs. Furthermore, quality of care is supposed to be enhanced by selective contracting (e.g., volume standards for breast cancer treatment).

Patient experiences are not systematically assessed. Although progress has been made, public reporting on quality of care and provider performance is still in its infancy in the Netherlands. Patients may report individual experiences with healthcare providers and institutions in any sector to the website Zorgkaartnederland.nl on a voluntary basis. Furthermore, several websites provide information about institutions (including hospitals and nursing homes) and providers in any sector, primarily based on quality indicators obtained from the National Quality Institute and the Dutch Healthcare Inspectorate.

What is being done to reduce disparities?

Every four years, variations in health accessibility are measured and published in the Dutch Health Care Performance Reports by the National Institute for Public Health and the Environment, focusing on socioeconomic differences including ethnicity and education. Geographic or regional variation is not measured consistently. Socioeconomic health disparities are considerable in the Netherlands, with up to seven years' difference in life expectancy between the highest and lowest socioeconomic groups. Smoking is still a leading cause of death. Although monitored by the National Institute for Public Health and the Environment (part of the Ministry of Health), the government does not have specific policies to overcome health disparities. In 2013, government decided to include diet advice and smoking cessation programs in the statutory benefits package.

What is being done to promote delivery system integration and care coordination?

A bundled-payment approach to integrated chronic care is applied nationwide for diabetes, COPD, and cardiovascular risk management. Under this system, insurers pay a single fee to a principal contracting entity – the care group (see above) – to cover a full range of chronic disease services for a fixed period. The bundled-payment approach supersedes traditional health care purchasing for the condition and divides the market into two segments – one in which health insurers contract care from care groups, and another in which care groups contract services from individual providers, each with freely negotiable fees [5]. The role

of district nurses is currently being strengthened to better coordinate care and help reach vulnerable populations.

Over the last years, a number of pilot studies across the Netherlands have been initiated to improve integration and coordination, primarily focusing on health and lifestyle improvement, population management and administrative simplification. These initiatives have been met with mixed success.

What is the status of electronic health records?

Virtually all general practitioners have a degree of electronic information capacity – for example, they use an electronic health record (EHR) and can order prescriptions and receive lab results electronically. At present, all hospitals have an electronic health record. Providers must allow patients access to their own files upon request, but few hospitals have standard online access options for patients. Electronic records for the most part are not nationally standardized or interoperable between domains of care. In 2011 legislation to install a national electronic health record system failed in congress. Since then, integration of different EHR systems between hospitals and between hospitals and other providers has been left to the field. In 2011, hospitals, pharmacies, after-hours general practice cooperatives, and organizations representing general practitioners set up the Union of Providers for Health Care Communication (De Vereniging van Zorgaanbieders voor Zorgcommunicatie), responsible for the exchange of data via an IT infrastructure named AORTA; data are not stored centrally. Patients must approve their participation in this exchange and have the right to withdraw. Aorta uses unique provider identification numbers and patient social security numbers, in oversight of the government agency Central Healthcare Information and Occupation Access Point. In practice, use of this system is limited. Other initiatives have focused on improving data exchange. For example, MedMij is a private organization that develops a package of standards and agreements that ensure that portals, provider systems, and apps can be linked to safely exchange information.

How are costs contained?

The main approach to controlling costs relies on market forces while regulating competition and improving efficiency of care. In addition, provider payment reforms, including a shift from a budget-oriented reimbursement system to a performance- and outcome-driven approach, have been implemented. In light of the global financial crisis, additional activities have been undertaken in order to contain costs. Since 2012, healthcare spending has declined from 10.9% to 10.5% of GDP.

In 2011, an agreement signed by the minister of health, all health care providers, and insurers set a voluntary ceiling for the annual growth of spending on hospital and mental care. When overall costs exceed that limit, the government has the ability to control spending via generic budget cuts. Because of the sector agreements, it has been argued that hospitals and insurers de facto negotiate lump sum contracts with budget ceilings as the most

important provision. The agreement included an extra 1 percent spending growth allowance for primary care practices in 2014 and 1.5 percent in 2015–2017, provided they demonstrate that their services are a substitute for hospital care. These agreements were prolonged for one year until the end of 2018, and the current Ministry of Health has successfully negotiated new agreements that will expire at the end of 2022.

The pharmaceutical sector is generally considered to have contributed significantly to the decrease in spending growth. Average prices for prescription drugs declined in 2014, although less than in previous years, with reimbursement caps for the lowest-price generic contributing to the decrease in average price. Reimbursement for expensive drugs has to be negotiated between hospital and insurer; there is some concern, however, that this and other factors may limit access to expensive drugs in the near future.

Health technology assessment is gaining in importance and is used mainly for decisions concerning the benefit package and the appropriate use of medical devices. The management of the basic benefit package also contributes to cost-containment. Based on the advice by the Healthcare Institute, the Minister has negotiated lower prices with the manufacturers for a range of expensive drugs. The Dutch health minister has formulated an ambitious policy proposal aiming in part to limit the pharmaceutical industry's power over drug pricing. During the Dutch European Union presidency in 2016, the topic was successfully put on the European Union agenda but the effectiveness of the proposed policies remains to be seen.

The annual deductible, which accounts for the majority of patient cost sharing, has more than doubled between 2008 and 2018, from EUR170 (USD210) to EUR385 (USD475). There are some worries that this increase has led to greater numbers of people abstaining from or postponing needed medical care.

Cost containment is most severe in long-term care. Since 2013, people with lower care needs are no longer entitled to residential care. In addition, the devolution of services to the municipalities as a result of the 2015 Long-Term Care Act was accompanied by substantial cuts to the available budgets (on average almost 10%).

The Federation of University Medical Centers has recently started a program aimed at reducing lower-value services. In addition, the Dutch Federation of Medical Specialists launched the "Dutch Choosing Wisely" campaign, which is also aimed at reducing lower-value services.

What major innovations and reforms have been introduced?

Long-term care, including home care, was under separate legislature (the Exceptional Medical Expenses Act) up to 2015. In 2015, the major reform placed residential long-term care under the newly created Long-Term Care Act, and transferred home care to the Health Insurance Act (medical home care and home nursing care) and Social Support Act (ancillary home services). The reform program's main goals were to guarantee fiscal sustainability and universal access in the future and to stimulate greater individual and social responsibility. The devolution of services to the municipalities as a result of the 2015 Long-Term Care Act was accompanied by substantial cuts to the available budgets (on average almost 10%).

In 2015–2016, initial budget reductions have been retracted, and future budget increases

worth €2.1 billion are set aside by the government to alleviate fiscal stress in nursing homes [6].

In curative health care, market reform and regulated competition remain somewhat controversial. The government, determined to continue stimulating competition between insurers and providers, undertook some measures to that effect, such as requiring insurers and providers to assume greater financial risk. Affordability and the accessibility of expensive drugs have rapidly become prominent issues [7].

As of the date of this report, the Health Insurance Act of 2006 has undergone two evaluations [8]. The latest evaluation pointed to an imbalance of power, with providers having an advantage over insurers.

References

1 Organisation for Economic Co-operation and Development (OECD), OECD.Stat., https://stats.oecd.org/Index.aspx?DataSetCode=SNA_TABLE4, accessed May. 8, 2018. Please note that, throughout this profile, all figures in USD were converted from euros at a rate of about 0.81 euros per USD, the purchasing power parity conversion rate for GDP in 2017 reported by OECD (2018) for the Netherlands.

2 Kroneman, M., et al. (2016). "Health systems in transition." Health 18(2).

3 W. Schäfer, M. Kroneman, W. Boerma et al., "The Netherlands: Health System Review," in Health Systems in Transition (European Observatory on Health Systems and Policies, 2010).

4 P. C. Smith, A. Anell, R. Busse et al., "Leadership and Governance in Seven Developed Health Systems," Health Policy, June 2011 106(1):37–49.

5 J. N. Struijs and C. A. Baan, "Integrating Care Through Bundled Payments – Lessons from the Netherlands," New England Journal of Medicine, March 17, 2011 364(11):990–91.

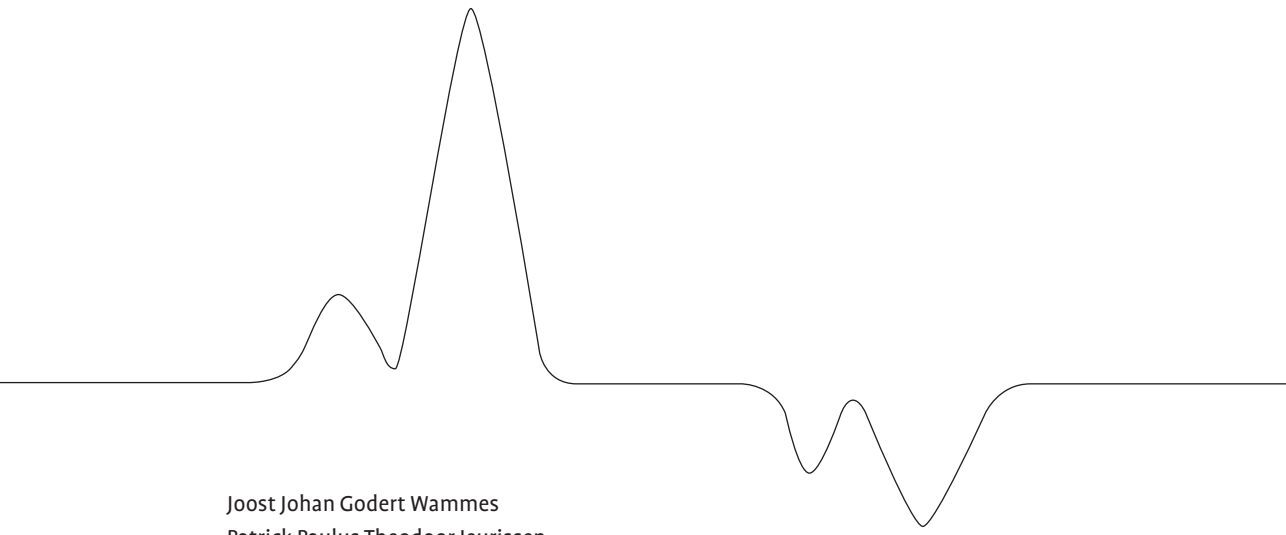
6 J. A. Maarse and P. P. Jeurissen, "The Policy and Politics of the 2015 Long-Term Care Reform in the Netherlands," Health Policy, March 2016 120(3):241–45.

7 H. Maarse, P. P. Jeurissen, D. Ruwaard, "Results of the Market-Oriented Reform in the Netherlands: A Review," Health Economics, Policy and Law, April 2016 11(2):161–78.

8 Plexus, K. (2014). Evaluatie Zorgverzekeringswet. Eindrapportage september; van de Ven, W., Schut, E., Hermans, H., de Jong, J. D., Van der Maat, M., Coppen, R. C., Groenewegen, P., and Friele, R. (2009). "Evaluatie Zorgverzekeringswet en Wet op de zorgtoeslag."

CHAPTER 3

Is the role as gatekeeper still feasible? A survey among Dutch general practitioners



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Fam Pract. 2014 Oct;31(5):538-44.

Abstract

Background In the 2012 International Health Policy Survey by the Commonwealth Fund, 57% of Dutch general practitioners (GPs) indicated that Dutch patients receive too much healthcare. This is an unexpected finding, given the clear gatekeeper role of Dutch GPs and recent efforts strengthening this role.

Objectives The study aims to explore where perceived overuse of care prevails and to identify factors associated with too much care at the entry point of Dutch healthcare.

Method An American survey exploring perceptions of the amount of care among primary care providers was modified for relevance to the Dutch health system. We further included additional factors possibly related to overuse based on twelve interviews with Dutch GPs. The survey was sent to a random sample of 600 GPs.

Results Dutch GPs (N=157; response rate 26.2%) indicated that patients receive (much) too much care in general hospitals, in primary care, in GP cooperatives as well as in private clinics. The Dutch responding GPs showed a relatively demand-satisfying attitude, which contributed to the delivery of too much care, often leading to deviation from guidelines and professional norms. The increasing availability of diagnostic facilities was identified as an additional factor contributing to the provision of unnecessary care. Finally, funding gaps between primary care and hospitals impede cooperation and coordination, provoking unnecessary care.

Conclusion Our results – most notably regarding the demand-satisfying attitude of responding GPs – call into question the classical view of the guidance and gatekeeper role of general practitioners in the Dutch healthcare system.

Background

57% of Dutch general practitioners (GPs) believe patients receive (much) too much medical care. This was one of the main findings of the International Health Policy Survey (IHP), a longitudinal international comparative research study in Western countries exploring the experiences of general practitioners (GPs) with care. Of the ten countries, only Germany scored slightly higher (59%), while on the contrary in New Zealand 39% of GPs felt that patients actually received too little care [1].

The general consensus is that a well-functioning primary care system provides stepped care: right care at the right place, on the right time, balancing quality and costs. The GP provides care at relatively low cost and avoids costly hospital care [2]. For that reason, in various countries policy is aimed at further strengthening the gatekeeper role of primary care. Dutch examples are the introduction of GP cooperatives for after-hours care run by general practitioners [3], the increased availability of diagnostic facilities in general practice, and the promotion of integrated care for the chronically ill through bundled payments [4]. In Germany, a nationwide primary care-based and physician-sustained disease management program has improved quality of care, while also curbing costs [5]. In the United States patient-centred medical homes are considered to be the most popular primary care delivery innovation, capable of improving quality of care while reducing costs [6]. Recent research indicates that this model also holds a promise for other countries [7].

Meanwhile, a Dutch health policy study suggested an erosion of the role of the general practitioner: many patients receive specialized medical care without consulting the general practitioner beforehand [8]. We have known for some time that large differences exist in care between GP practices, with implications for the effectiveness of GP care [9]. A recent study in 31 European countries showed that a strong primary care system is not only associated with better population health, but also with higher health spending [10].

To summarize, many countries are strengthening their primary care system, while at the same time the effectiveness and consistency of these systems is sometimes being questioned. The findings of the IHP survey mentioned above draw attention to a remarkable observation: how do we explain the fact that Dutch GPs experience so much excess care while working in a health care system with a clear gatekeeper? To find out, we conducted an exploratory study among Dutch GPs aiming to 1) understand where this perceived care prevails; 2) identify factors that are associated with too much care at the entry point of Dutch healthcare. In doing this, we aim to test assumptions supporting the gatekeeper system and further strengthening of this gatekeeper system. Therefore, we asked the respondents to assess the perceived amount of care by sector and type. Secondly, we used clinical cases to gain information about practice patterns and to identify factors/motivations for choice of policy in that specific clinical case. Furthermore, we used several propositions describing factors possibly related to overuse and asked the respondents for opinion.

Methods

We used a previously conducted American questionnaire exploring perceptions about the amount of care among primary care providers, as a starting point [11]. This questionnaire was translated into Dutch by a certified translation company. We subsequently included additional factors possibly related to overuse on the basis of a literature study and interviews with twelve Dutch GPs and adapted the questionnaire to the Dutch situation. The aim of the interviews was to describe cases of overuse and to identify factors possibly related to overuse. The interviews were thematically analyzed by two independent researchers, to extract the relevant factors driving overuse in the Dutch healthcare system. A third researcher was consulted in case of disagreement. Ultimately, 17 questions were taken from the American questionnaire (eg. a clinical case or questions concerning incidental findings which for which we changed the clinical case to be applicable in the Netherlands), while the remaining 19 questions were formulated on the findings of our interviews. In short, the questionnaire consisted of items considering perceived amount of care by sector and type, and factors concerning or related to practice policy, the gatekeeper role, referrals, the role of the patient, diagnostics, awareness of costs, the health system, and other factors possibly related to too much care. Most questions were in the form of propositions describing a factor possibly related to overuse (with some in context of a clinical case), with a 5-point likert to agree/disagree or alike. The questionnaire was tested for consistency and comprehensibility through cognitive interviewing with a GP.

A random sample of 600 GPs, drawn from the NIVEL database 'Health professions', was invited to participate by means of an invitational letter. This letter was accompanied by a written questionnaire and a postage-paid business reply envelope. Two weeks after the first mailing, non-responding GPs received a reminder in the form of a postcard. Four weeks after the first dispatch, non-responding GPs received a new copy of the questionnaire and again a freepost return envelope. The results of the questionnaires were analyzed using SPSS version 20. Respondents who did not reply to some of the questions were still included in the analysis; questions without an answer were considered missing. Below, the most salient results will be presented.

Results

Respondent characteristics

A total of 157 GPs (response rate = 26.2%) completed the questionnaire, 100 general practitioners (16.7%) indicated that they did not want to participate. Our sample was representative for the entire Dutch GP population (Table 1), with a slight over-representation of older male GPs as well as GPs in paid employment. Almost 80% indicated that they are self-employed and about two-thirds of the respondents reported having a working week of more than 40 hours.

TABLE 1 Characteristics of respondents and national GP population

		Respondents (N=157)	Dutch GP population[1] (N=8831)
Gender	Male (%)	60.5%	57.5%
	Female (%)	39.5%	42.5%
Age (years)	Average	51.2 ♂54.5 ♀45.8	 ♂51 ♀44[2]
	< 35 (%)	3.9%	6.6%
	35-44 (%)	21.4%	29.0%
	45-54 (%)	29.9%	32.5%
	55-64 (%)	44.2%	30.9%
	65+ (%)	0.6%	1.0%
Primary practice setting	Solo practice (%)	22.5%	25.7%
	Two-person practice (%)	29.8%	37.9%
	Group practice (%)	29.1%	36.4%
	Health care centre (%)	17.2%	
	Other (%)[3]	1.4%	
Ownership status	Self-employed (%)	78.8%	88.9%
	Paid employment (%)	21.2%	11.1%
Percentage of patients in the practice older than 65	Under 10% (%)	7.2%	Unknown
	10 to 25% (%)	46.4%	Unknown
	26 to 50% (%)	45.1%	Unknown
	Over 50% (%)	1.3%	Unknown

[1] Source: www.nivel.nl/databank (All Dutch GPs are included in this databank.)
[2] Source: Nivel. Cijfers uit de registratie van huisartsen. Peiling 2010.
[3] The results under "other" usually contained digressions on one of the alternatives.

Amount of care by sector and type

A very large majority (81.4%) indicated that in their perception, patients in the Netherlands receive (much) too much care (Table 2, in the rest of the article we do not repeat the adjectives (much) too much, very or strongly used in our 5-point likerts). Focusing on type of care, a few highlights can be observed: over 80% of respondents felt that too much care is delivered in private clinics, at GP cooperatives and in hospitals. Moreover, respectively 35.5% and 36.1% of the respondents indicated that patients receive much too much care at the GP cooperative and in private clinics. More than half of the GPs (58.2%) considered that too much care was delivered by general practitioners themselves. In contrast, 63.2% of respondents indicated nursing and residential care homes as settings where patients receive too little care. Only the amount of palliative care was relatively often perceived as being just right (67.3%).

Almost all respondents (90.9%) felt that patients received too much diagnostic care. Also, medical treatment (78.7%) as well as monitoring and follow-up (48.7%) are provided too much according to the participating GPs.

TABLE 2 Opinion of Dutch GPs on amount of care patients received (by sector and type).

	Much too little	Too little	Just about right	Too much	Much too much
General (IHP-question)[1]	0.0%	0.7%	17.9%	71.5%	9.9%
Sector					
Private clinics[2,4]	0.0%	1.6%	13.9%	48.4%	36.1%
GP cooperative[2]	0.0%	1.9%	13.5%	49.0%	35.5%
Hospital[2]	0.0%	4.5%	11.0%	69.5%	14.9%
Primary mental health care2	0.0%	34.7%	47.9%	14.6%	2.8%
Secondary mental health care[2,4]	0.7%	40.3%	38.1%	17.2%	3.7%
General practitioner care[2]	0.0%	5.2%	36.6%	56.9%	1.3%
Home care[2]	2.6%	37.3%	50.3%	9.2%	0.7%
Nursing and residential care homes[2]	6.1%	57.1%	30.6%	6.1%	0.0%
Type					
Diagnostics[3]	0.0%	0.6%	8.4%	74.0%	16.9%
Medical treatment[3]	0.0%	0.0%	22.2%	69.9%	7.8%
Monitoring and follow-up[3]	0.0%	9.7%	41.6%	43.5%	5.2%
Prevention[3]	4.6%	48.7%	23.0%	19.7%	3.9%
Rehabilitation[3]	0.7%	43.1%	52.1%	3.5%	0.7%
Nursing and care[3]	2.0%	57.8%	38.8%	1.4%	0.0%
Palliative care[3]	0.0%	32.0%	67.3%	0.7%	0.0%

[1] Question from Commonwealth Fund IHP survey: Thinking about all the medical care your patients receive – not just from you, but from all their providers, including specialists – what is your opinion about the amount of medical care they receive? Is it…?
[2] Thinking about all the medical care your patients receive, what is your opinion about the amount of care they receive at the ….
[3] Thinking about all the medical care your patients receive, what is your opinion about the amount of …….. care they receive.
[4] 22 and 35 respondents chose the option 'Do not know' for secondary mental health care and private clinics respectively.

Clinical cases and variation in treatment by GP

CASE 1 A patient of yours (60-year-old man) has well-controlled hypertension. This is his only medical problem.

To the question 'In general, how frequently do you schedule routine follow-up visits? Every ... months" the largest group responded every 6 months (42.5%), followed by every 12 months (30.1%) and every three months (24.8%) (N = 153).

We presented the respondents with the additional cases 2 and 3 (see boxes and tables 3 and 4). We asked them to what extent specific factors determined their policy. The three cases showed that there is a large variation in practice among the participating GPs. The GPs explained that their choice depended on the degree of anxiety and awareness of the patient and the degree to which the patient accepts the given explanation. Almost all respondents (more than 80%) indicated that this somewhat or substantially played a role in their choice of policy. Their choice was further motivated by doing what was indicated "on clinical grounds" (62.4%) and "doing what the patient expects him to do" (55.6%). It is also notable that there seems to be a division among the participating GPs: 42.9% indicated that the clinical indication largely determined their choice, while on the other hand 37.7% stated that the clinical indication did not influence their decision.

CASE 2 A mother contacts the practice by phone about her ten-year-old daughter. She has been coughing for two days and has a rise in temperature (38.5°) since last night. The daughter does not feel like eating, drinks well but started coughing heavily again last night. Her mother would like to have a consultation this afternoon. The assistant tries to give advice and explain to her that it would not seem to be necessary to visit the practice. Still, the mother continues to ask for a consultation and the assistant would like to confer with you.

CASE 3 For many years, your patient has suffered from chronic daily headaches. This worries him considerably. You know the patient well, he often visited your practice for these complaints. You have never been able to find out the cause. The last medication you prescribed also had no effect. The patient is distraught and asks for a CT scan or MRI of the head to be made.

TABLE 3 What would be your policy with regard to case 2?

Policy	
Tell the assistant that the proposed policy (the assistant informs the mother that it is not necessary to come to the practice) is all right and that the demand for office visits will not be honored.	5.7%
You ask the assistant to call the mother and ask her to wait for a while. If the fever persists for more than three days, she can phone in the morning for an appointment.	35.7%
You offer the mother a consultation by telephone.	22.3%
You let the mother and daughter come to the practice.	36.3%

TABLE 4 What would be your policy with regard to case 3?

Policy	
You try to calm the patient and explain that a CT scan or MRI for these complaints is not useful.	21.7%
You request a CT scan or MRI for him.	3.9%
You refer the patient to a neurologist.	74.3%

Patient-provider relationship factors

Nearly all (> 90%) respondents indicated that patients experience health care as a right, and that this fact leads to unnecessary care. A comparable proportion indicated that patients have a strong need for an explanation and certainty, and that this also leads to unnecessary care. Two thirds (66.1%) stated that when patients really wanted to be referred, they would go along with this as they prevailed to maintain the relationship with the patient.

Provider decision-making and clinical guideline issues

Pancreatic lipomatosis in an ultrasound for possible cholelithiasis is an example of incidental findings in diagnostics. To the question "How often are you faced with such incidental findings?" 29.4% of the respondents answered often and 6.5% very often. It is notable that one-third (32.2%) of the respondents usually and 4.6% almost always request additional tests to clarify incidental findings. Only a minority (38.0%) indicated that they do not bother to deviate from the written recommendation of the radiologist when confronted with incidental findings. Most GPs indicated that they felt obliged to follow the recommendation (23.3%) or deviate from the recommendation only in exceptional circumstances (38.7%). Additionally, more than two-thirds (70.9%) found that the availability of diagnostic tools (ECG/spirometry) at the practice leads to more investigations, as opposed to these investigations having been requested. Moreover, according to 61.6% of the respondents, some guidelines prescribe so many monitoring tests that they feel they request these required tests unnecessarily.

Issues related to the relationship between primary care and other sectors

According to the respondents, a variety of factors may increase or decrease the number of referrals. A lack of time at the moment of referral and fear of making mistakes led to an increased number of referrals (more than 60% of the respondents). According to 70.9% of the participating GPs, patients are reassured more quickly when he or she refers increasing the number of referrals. More than half (54.1%) indicated that it takes a lot of time and effort to convince a patient that an additional investigation is not beneficial. This further increased the number of referrals.

The fact that patients easily receive hospital care without a referral from a general practitioner leads to unnecessary care, according to 64.3% of the GPs. Almost 80% of respondents indicated that some patients prefer the GP cooperative as an alternative for the regular practice-based primary care and that this leads to unnecessary care. Over 80% of respondents considered that insurers reimburse care in hospitals that could be provided by the GP, which provoked unnecessary care. 70% thought that insurers could more actively guide providers to reduce unnecessary care. 72.4% indicated that funding gaps between primary care and hospitals impede cooperation and coordination, which provoked unnecessary care.

Discussion

According to the 26% of invited Dutch GPs who responded to this survey, patients receive too much care in general hospitals, in primary care, in GP cooperatives as well as in private clinics. The Dutch responding GPs' demand-satisfying attitude and the increased availability of diagnostic facilities most saliently contribute to the provision of perceived excess care at the entry point of care in the Netherlands. Also misaligned incentives induce that Dutch responding GPs do not sufficiently pick up the gatekeeper's role. Below, these findings are discussed more elaborately.

Our results show that responding practitioners find it difficult to deny demanding patients access to further care, even if they think treatment is unnecessary from a medical point of view. This creates an image of responding GPs acting in a demand-satisfying way in their referrals and treatment decisions. It is likely that this contributes to the perceived amount of overdiagnosis and overtreatment.

The demand-satisfying attitude of the responding GPs puts into question the classic, possibly simplified, image of the 'gatekeeper' impeding access to expensive unnecessary hospital care. According to 84.1% of the respondents, too much care is provided at hospitals, an indication that the gatekeeper system, originally meant to be a gateway to secondary care, is working suboptimal. A recent Dutch study showed a threefold variation in referral rates to medical specialists between GPs. This variation was driven by the physician practice pattern, not by the patient case mix [12]. Undoubtedly, this variation leaves ample room for improvement, although the optimal level of referral is unknown. A myriad of policy options is available to reduce referrals, such as tightening or more explicitly defining the criteria for referral, implementing (multidisciplinary) guidelines, increasing conversation and collaboration between primary and secondary care or benchmarking GPs on referral rates [12-15].

The possibility of bypassing the general practitioner in favour of hospital care (for example via the emergency department), as well as budget gaps between primary care and hospitals and the absence of guiding insurers impede general practitioners in maintaining their role of gatekeeper. Our results show that responding GPs themselves are prepared to avoid perceived unnecessary hospital care – versus reducing perceived unnecessary care in primary care – yet that the preconditions at the level of the health system do not meet. Bundled payments or medical specialist consultation at primary care practice may in theory (partly) overcome this problem [16].

The three cases illustrate that there is probably a large variation in practice among Dutch responding GPs. Such variation suggests that some patients receive suboptimal care and that there is ample room for improvement. Our findings indicate that Dutch responding GPs are not determined to their role of commissioners of care. Moreover, responding GPs admit to providing a lot of unnecessary care themselves. The combination of a demand-satisfying attitude of the Dutch responding GPs, with consumerism among patients (patients perceive health care as their right), drives this perception. Shared decision making may be a feasible strategy to address both factors. The evidence-based source Clinical evidence estimates that only a minority of treatments is 'beneficial' (11%) or 'likely to be beneficial' (24%). The remaining treatments were classified at best as 'a trade-off between benefits

and harms' (7%) to having an 'unknown effectiveness' (50%) [17]. Thus, in many instances an evidence base may not be able to provide the best alternative. Rather, two or more medically acceptable alternatives may exist, whereby the choice should be dependent on the patients' preferences and the possible harms and benefits of each alternative [18]. Research shows that when patients are better informed they tend to opt for a more conservative approach [19]. An example of this is the study done by Fleuren et al, who showed that the implementation of a shared care guideline for lumbosacral radicular syndrome reduces unnecessary early referrals [20].

According to responding GPs, the availability of (access to) new diagnostic facilities in primary care leads to added risk of accidental discoveries and follow-up treatments, but also to unnecessary diagnostics itself. Therefore, this seems to contribute to perceived unnecessary care. Our findings raise the question of how to combine the increased possibilities for diagnosis and treatment at primary care with a prudent use. Due to the increasing possibilities for diagnosis and treatments, supply-induced demand might become a major theme in primary care as well.

Remarkably, our study showed that responding GPs sometimes question the necessity of care provided and requested at GP cooperatives (which is actually care delivered by responding GPs themselves). In 2006, Giesen et al [21] found that more than three quarters of all contacts at GP cooperatives did not concern urgent problems, which may explain the perceived amount of excess care we found. Both our study and the study by Giesen et al raise the question how to practically shape the GP cooperatives. Both observations concerning diagnostic facilities and GP cooperatives are relevant, since these are actively encouraged in the Netherlands and in many OECD-countries, one of their objectives being the reduction of unnecessary care.

Finally, we confirmed the finding of Sirovich et al [11] that due to a lack of time responding GPs practice in a more active style concerning ordering diagnostic tests and referrals, although malpractice concern and clinical performance measures play a less prominent role in the Netherlands, as opposed to the USA. In the Netherlands the average numbers of inhabitants per GP is 2300, which is relatively high. This may explain the working pressure and active practice style.

Limitations

The participating GPs were slightly older, did less frequently work in a solo practice, and were more often than the national average in paid employment. Given the difference in score on the IHP-question (81% in our study compared to 57% in the IHP-survey [1]), response bias may have played a role in this study in the sense that more critical GPs may have been more likely to respond. Even so, this will not necessarily affect the validity of the identified factors that we found to be related to excess or unnecessary care. Moreover, non-response studies among physicians have shown no or minimal amounts of response bias, suggesting that physician surveys are more resilient to non-response than other types of surveys [22]. Overall, we conclude that the low response rate may represent some response bias but given the explorative nature of the study and the bold statements made by a substantial group of GPs, the results justify further research.

Conclusion

This study shows that, according to Dutch responding GPs, a lot of unnecessary care is delivered in hospitals, GP cooperatives and in private clinics. According to the responding GPs the demand-satisfying attitude of general practitioners contributes to perceived unnecessary care, as does the increased availability of diagnostics. The assumption that the costs of additional investments in primary care will be automatically paid back by reducing unnecessary care at hospitals needs to be further investigated. The various roles of the GP – gatekeeper, patient navigator, therapist and navigator – are of interest in this. Shared decision making has most potential in addressing both the demand-satisfying attitude of GPs and consumerism among patients. However, questions remain regarding the potential impact of such a strategy and more research on shared decision making and alternatives is needed, because it is still in stage of infancy/a novel phenomenon. Our results indicate that further discussion and exploration by GPs and policy makers about the complicated and sometimes unintended effects of strengthening primary care and its interactions with unnecessary care may be fruitful. Supply-induced demand does not stop beyond medical specialists; primary care doctors are 'vulnerable' to it as well.

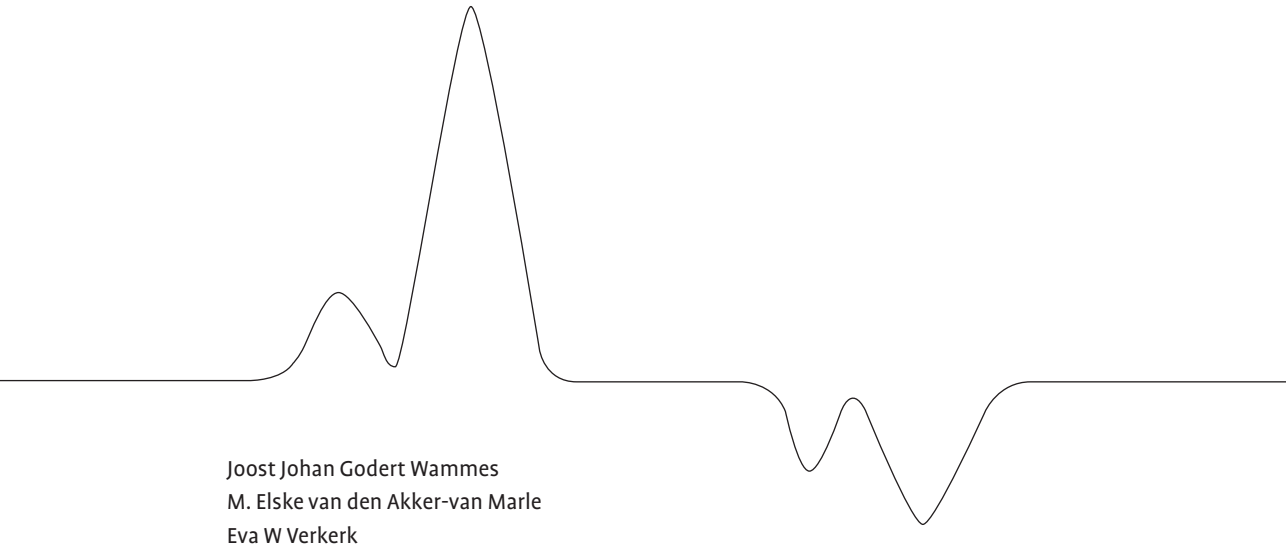
References

1 Schoen C, Osborn R, Squires D, et al. A survey of primary care doctors in ten countries shows progress in use of health information technology, less in other areas. Health affairs. Dec 2012;31(12):2805-2816.
2 Starfield B, Shi L, Macinko J. Contribution of primary care to health systems and health. The Milbank quarterly. 2005;83(3):457-502.
3 Thijssen WA, Wijnen-van Houts M, Koetsenruijter J, Giesen P, Wensing M. The impact on emergency department utilization and patient flows after integrating with a general practitioner cooperative: an observational study. Emergency medicine international. 2013;2013:364659.
4 NZa. Consultatiedocument Eerstelijnsdiagnostiek. Goede beeldvorming beter in beeld. 2011.
5 Stock S, Drabik A, Buscher G, et al. German diabetes management programs improve quality of care and curb costs. Health affairs. Dec 2010;29(12):2197-2205.
6 Hoff T, Weller W, DePuccio M. The patient-centered medical home: a review of recent research. Medical care research and review : MCRR. Dec 2012;69(6):619-644.
7 Faber M, Voerman G, Erler A, et al. Survey Of 5 European Countries Suggests That More Elements Of Patient-Centered Medical Homes Could Improve Primary Care. Health affairs. Apr 2013;32(4):797-806.
8 Plexus K. Onderzoek naar de effectiviteit van poortwachters in het Nederlandse zorgstelsel 2000 – 2010. 2012.
9 Cardol M, Dijk Lv, Jong Jd, Bakker Dd, Westert G. Huisartsenzorg: wat doet de poortwachter? : NIVEL. RIVM. ;2004.
10 Kringos DS, Boerma W, van der Zee J, Groenewegen P. Europe's strong primary care systems are linked to better population health but also to higher health spending. Health affairs. Apr 2013;32(4):686-694.
11 Sirovich BE, Woloshin S, Schwartz LM. Too Little? Too Much? Primary care physicians' views on US health care: a brief report. Archives of internal medicine. Sep 26 2011;171(17):1582-1585.
12 van Dijk C, Korevaar J, de Jong J, Koopmans B, van Dijk M, de Bakker D. Ruimte voor substitutie? Verschuivingen van tweedelijns- naar eerstelijnszorg. NIVEL;2013.
13 Iacobucci G. GPs put the squeeze on access to hospital care. Bmj. 2013;347:f4432.
14 Katz MH. How can we know so little about physician referrals? Archives of internal medicine. JAN 23, 2012 2012;172(2).
15 Dijkstra R, Brugge At. De poortwachtersrol in het licht van toekomstbestendige hoogwaardige Gezondheidszorg. Amsterdam: Coincide;2013.
16 Struijs JN, Baan CA. Integrating care through bundled payments--lessons from The Netherlands. The New England journal of medicine. Mar 17 2011;364(11):990-991.
17 What conclusions has Clinical Evidence drawn about what works, what doesn't based on randomised controlled trial evidence? http://clinicalevidence.bmj.com/x/set/static/cms/efficacy-categorisations.html. Accessed 19 februari, 2014.
18 Stiggelbout AM, Van der Weijden T, De Wit MP, et al. Shared decision making: really putting patients at the centre of healthcare. Bmj. 2012;344:e256.

19 Stacey D, Legare F, Col NF, et al. Decision aids for people facing health treatment or screening decisions. The Cochrane database of systematic reviews. 2014;1:CD001431.
20 Fleuren M, Dusseldorp E, van den Bergh S, et al. Implementation of a shared care guideline for back pain: effect on unnecessary referrals. International journal for quality in health care : journal of the International Society for Quality in Health Care / ISQua. Oct 2010;22(5):415-420.
21 Giesen P, Franssen E, Mokkink H, van den Bosch W, van Vugt A, Grol R. Patients either contacting a general practice cooperative or accident and emergency department out of hours: a comparison. Emergency medicine journal : EMJ. Sep 2006;23(9):731-734.
22 Flanigan T, McFarlane E, Cook S. Conducting Survey Research among Physicians and other Medical Professionals – A Review of Current Literature. Proceedings of the Survey Research Methods Section; 2008.

CHAPTER 4

Identifying and prioritizing lower value services from Dutch specialist guidelines and a comparison with the UK do-not-do list



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Abstract

Background The term 'lower value services' concerns healthcare that is of little or no value to the patient and consequently should not be provided routinely, or not be provided at all. De-adoption of lower value care may occur through explicit recommendations in clinical guidelines. The present study aimed to generate a comprehensive list of lower value services for the Netherlands that assesses the type of care, and associated medical conditions. The list was compared with the NICE do-not-do list (United Kingdom). Finally, the feasibility of prioritizing the list was studied – to identify conditions where de-adoption is warranted.

Methods Dutch clinical guidelines (published from 2010-2015) were searched for lower value services. The lower value services identified were categorized by type of care (diagnostics, treatment with and without medication), type of lower value service (not routinely provided or not provided at all) and ICD10-codes (international classification of diseases). The list was prioritized per ICD10-code, based on the number of lower value services per ICD10-code, prevalence and burden of disease.

Results A total of 1366 lower value services was found in the 193 Dutch guidelines included in our study. Of the lower value services 30% covered diagnostics, 29% related to surgical and medical treatment without drugs primarily and 39% related to drug treatment. The majority (77%) of all lower value services was on care that should not be offered at all, whereas the other 23% recommended on care that should not be offered routinely. ICD10-chapters that included most lower value services were neoplasms and diseases of the nervous system. Dutch guidelines appear to contain more lower value services than UK guidelines. The prioritization processes revealed several conditions – including back pain, COPD and ischaemic heart diseases – where lower value services most likely occur and de-adoption is warranted.

Conclusion In this study, a comprehensive list of lower value services for Dutch hospital care was developed. A feasible method for prioritizing lower value services was established. Identifying and prioritizing lower value services is the first of several necessary steps in reducing them.

Background

Quality of healthcare is reflected by "the degree to which health services for individuals and populations increase the likelihood of desired health outcomes and are consistent with current professional knowledge" [1]. Conform this definition, evidence-based medicine means that good medical practices are replaced by better ones when robust scientific evidence becomes available and practices that are outdated or proven invaluable to patients will be de-adopted. This ideal world is in sharp contrast with current medical practice [2,3].

Current practice is not always high-value or evidence-based. Lower value or lower quality of care may either be classified into misuse, overuse or underuse of health care services [4]. The focus of this paper is overuse; which occurs when a health care service is provided under circumstances in which its potential for harm exceeds the possible benefit [4]. In our study we also include (cost-)ineffective care, inappropriate timing of care or care not in line with the patients' wishes as lower value services. Many questions remain about the size of the problem. However, scientific literature suggests that overuse represents between 10% and 30% of provided services, of which a part is lower value care, resulting in worse outcomes including death and unnecessary costs [2,3,5]. We consider these services as lower value services, because they have no net value for the patient and de-adoption – a substantial reduction of providing or using the service in daily medical practice – is warranted.

During the last decade, efforts have been undertaken to de-adopt lower value services. UK's National Institute for Health and Care Excellence (NICE) started working on de-adoption in 2005 [6] which resulted in the 'do-not-do list' [7]. In the US, the National Physician Alliance started developing "Top Five" lists since 2009 and initiated the Choosing Wisely initiative in 2012 [8]. Australian activities were centered around the Medicare Benefits Schedule [9]. The basis of these programs is usually a (long) list of lower value services and sometimes a prioritization process to identify candidates for de-adoption [9,10].

The methods for creating these lists are diverse, and prioritization based on impact proves to be difficult. For example, Choosing Wisely lists varied widely in potential impact on daily care and spending; and specialist societies tended to list colleague specialties' services as lower value [8]. UK research has shown additional challenges, such as a lack of reliable evidence on the clinical merits of many services [11]. A prominent problem in overuse is that interventions which are high-value for a given subpopulation are inappropriately applied to other populations [12]. Candidate lists tend to be large and the potential gains in health and cost vary widely across lower value services. Therefore, as resources for de-adoption are limited, prioritization of lower value services for de-adoption is warranted.

To conclude, there is need for an objective approach to identify and prioritize lower value services for practical de-adoption [11]. This article describes the development of a list of lower value services identified from 193 Dutch clinical practice guidelines, published between 2010 and 2015. The list was developed with the aim to provide a comprehensive list of lower value services for Dutch hospital care. Furthermore, our list was compared with the NICE do-not-do list on several aspects, including types of care, and patient groups. Finally, the feasibility of prioritizing the list was studied. We hypothesized the prevalence of a disease and disease burden (a rationale for choice of criteria is given in the discussion) could serve as robust criteria for prioritization.

Methods

Development of lower value services list

Dutch guidelines contain specific recommendations to ensure that lower value care is not offered, or only applied to specific subpopulations or under limiting conditions. In the current study we identified these do-not-do recommendations. We have limited the analysis to the most recent and up to date guidelines published between January 2010 and May 2015 by the scientific societies, as Dutch guidelines are recommended to be revised every five years [13]. The guidelines were taken from a guideline database hosted by the Dutch Association of Medical Specialists (www.kwaliteitskoepel.nl) covering (mental) hospital care.

Firstly, we randomly selected eleven guidelines which were fully read by four researchers (SD, EV, JW and MEAM) to identify recommendations on care that should not be offered and care that should not be offered routinely. For each do-no-do recommendation identified, we listed whether the key term identifying the do-not-do recommendation was one of the search terms applied by NICE in the 'do not do' study (for example, 'discontinued', 'should not', 'do not' [14]) or a new term that should be added (e.g. 'omit'). Recommendations that focused on too little use of care (underuse) were not included. For example: "Restraint is not necessary when starting opioids and will lead to a substantial deterioration in quality of life by the experienced severe shortness of breath" (Guideline Palliative care for people with COPD). Finally, recommendations that focus on organization of care were not included. For example, "It is not recommended that professionals who have no experience with patients / offenders with antisocial personality (disorder) address the issue of the committed violence" (Guideline Domestic violence in children and adults). A fifth researcher (RBK) was consulted in case of no consensus.

Furthermore, the specific section of the guideline in which the do-not-do recommendation was written was identified. The standard format of guidelines contains five sections: clinical question, recommendations, substantiation, considerations and justification. As in the first five guidelines all the recommendations were found in the sections 'recommendations' and 'considerations' of the guidelines, subsequently only these sections of the electronic/PDF copy of a guideline were searched with the terms from Table 1.

Another nine guidelines were independently screened by the four researches (SD, MEAM, EV and JW) to determine the inter-rater reliability. Inter-rater reliability was analyzed by calculating Fleiss' Kappa (k) for multiple raters [15].

Using this method, the other guidelines were screened (in total 193), and any ambiguities were discussed with another researcher until consensus was reached. When guidelines were not constructed according to the standard format and therefore did not contain the paragraphs with recommendations and considerations, they were fully screened. For each do-not-do recommendation identified we assessed whether the care should not be offered at all or should not be offered routinely to all patients and what type of care the recommendation was about: diagnosis, treatment without medication, treatment with medication, and a residual category.

Guidelines that have been published in English were screened with English terms. Patient versions of guidelines were not included and also addenda to guidelines with original publication date before 2010 were excluded.

TABLE 1 Shortlist search terms

Dutch [English translation]	English
Niet [Not]	Discontinue/discontinuation
Geen [No]	Not
Stop [Stop]	No
Onvoldoende [Insufficient]	Ineffective
Zelden [Seldom]	Uncertain
Alleen [Only]	Avoid
Kosten [Cost]	Rarely
Vermijd/Vermeden [Avoid]	Stop
Achterwege [Omit]	
Onnodig [Unnecessary]	
Afgeraden [Discourage]	
Ontraden [Dissuade]	
Staken/Gestaakt [Cease]	

Connection with International Classification of Disease, Tenth Edition (ICD10) code

The lower value services described in the do-not-do recommendations were provided with an ICD10-code by searching within the ICD10-encoding [16] on the condition in question. When necessary, additional information was sought in the guideline the lower value service originated from and/or Wikipedia. If the lower value service was related to two (or more) conditions the guideline topic was selected for the ICD10-coding. For example, the guidance 'European Guidelines on cardiovascular disease prevention in clinical practice' included the recommendation 'Beta-blockers and thiazide diuretics are not recommended in hypertensive patients with multiple metabolic risk factors increasing the risk of new-onset diabetes'. This recommendation was categorized to the ICD10-code for hypertensive diseases. If the patient population receiving the lower value service could not be related to an ICD10-code, for example in the case of prevention in a healthy population, then the ICD10-code of the disease prevented was chosen. For example, the lower value service 'Do not use throat swabs when investigating for possible meningococcal disease' concerns the population with suspected meningococcal disease. Since there is no ICD10-code for this population, the ICD10-code of meningococcal disease was chosen. Complex cases were discussed between two researchers until consensus was reached. ICD10-codes were then aggregated to ICD10-chapters, the highest level of categorization in ICD10.

Comparison with NICE do-not-do database

In the development of NICE guidelines, clinical practices were identified which should not be used at all or should not be used routinely. These practices have been collected in the "do-not-do database" [7]. NICE has made an Excel file of the database (dated September 29, 2015) available to us upon request. We compared the average number of do-not-do recommendations per NICE guideline with the Dutch number. Furthermore, for each recommendation from the NICE do-not-do database we assessed whether the care should

not be offered at all or should not be offered routinely and what type of care was concerned (diagnosis, treatment without medication, treatment with medication). Finally, the same procedure with respect to assigning ICD10-codes was followed.

Prioritization

Prioritization of conditions for further research on lower value services for de-adoption was done by aggregating the lower value services described in the do-not-do recommendations by ICD10-codes, as the data for prioritization were only available at this level of aggregation and not for individual lower value services. Per ICD10-code we identified prevalence estimates and disease burden as available in the Global Burden of Disease studies [17] (a detailed description of the methodology is given in appendix 1). Prioritization was based on the number of lower value services per ICD10-code, prevalence and burden of disease (expressed in Years Lived with Disabilities (YLD) and Disability Adjusted Life-Years (DALY)). Each criterion was categorized in four groups according to level. Per criterion the group with the highest levels was assigned four points. Subsequently, the ICD10-codes were prioritized by the sum of scores for the number of lower value services, prevalence, YLD and DALY (Method 1), the highest score (up to 16) indicating the highest priority for de-adoption. As we were interested in the impact of burden of disease measures on prioritization (both YLD and DALY reflect burden of disease) we omitted these criteria in sensitivity analyses, and the prioritization was repeated for the sum of the number of lower value services and prevalence (Method 2; maximum score 8). For the NICE do-not-do database the same prioritization was performed, using UK-specific data on prevalence, YLD and DALY. In appendix 1, a full description of the prioritization methodology is given.

Results

Descriptives Dutch list of lower value services

In total, 1366 lower value services were extracted from the 193 Dutch guidelines on (mental) hospital care, implying that each guideline contained on average 7.1 (modus=0; median=5; maximum=45) lower value services. Of these guidelines 29 did not contain any lower value services. The inter-rater reliability was 0.803 (Fleiss k), indicating a substantial agreement [18]. Table 2 shows the average number of lower value services per guideline between 2010 and 2015. The number of guidelines published in 2014 and 2015 was relatively low because of the ending of a subsidy program. The majority of lower value services was, if necessary after deliberation within the project group, successfully linked to an ICD10-code. In 98 cases (<8%), no ICD10-code could be assigned, predominantly because the recommendation was ambiguous concerning the patient group, or the patient group was insufficiently specific (e.g. ‘essentially, laparoscopic surgery does not require different fluid management than open surgery’).

TABLE 2 Number of lower value services per year in Dutch guidelines.

Year	Number of guidelines published	Number of lower value services	Average number of lower value services per guideline
2010	61	357	5.85
2011	41	249	6.07
2012	44	347	7.89
2013	35	312	8.91
2014	2	45	22.5
2015	6	59	9.83

Of the lower value services, 415 (30%) related to diagnostics, such as 'There is no place for FDG-PET in the detection of micro metastases' (guideline anus carcinoma, Dutch list). 399 lower value services (29%) related to non-drug treatment, such as 'The insertion of a pulmonary artery catheter (PAC) in case of acute heart failure is rarely needed' (guideline heart failure, both in Dutch list and NICE database). Finally, 527 lower value services (39%) related to drug treatment, such as 'Methotrexate is not recommended for hidradenitis suppurativa' (guideline acneiform dermatoses, Dutch list). The remaining 25 (2%) lower value services did not fit into these categories (e.g. vaccination or recommendations on referral and discharge procedures). The majority (77%) of all lower value services concerned care that should not be offered at all, whereas the other 23% recommended on care that should not be offered routinely.

Figure 1 shows the number of lower value services identified per ICD10-chapter. For the Dutch guidelines, 'neoplasms' and 'diseases of the nervous system' are the most frequent chapters, followed by 'symptoms, signs and abnormal clinical and laboratory findings – not elsewhere classified', 'diseases of the circulatory system', 'diseases of the musculoskeletal system and connective tissue' and 'mental and behavioral disorders'. Relatively few lower value services were found in ICD10-chapters 'external causes of morbidity and mortality', 'conditions originating in the perinatal period', and 'diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism'.

Comparison with NICE do-not-do recommendations

The database contained 188 guidelines in which 1006 do-not-do recommendations (lower value services) were found. The UK guidelines thus covered relatively few lower value services: on average 5.4 (modus=1; median=3; maximum=32) per guideline. UK guidelines covered slightly fewer lower value services related to diagnostics (28%) and non-drug treatment (25%), and relatively many lower value services related to drug treatment (46%). In addition, UK lower value services less likely described care that should not be offered at all (68%), whereas the other 32% recommended on care that should not be offered routinely. Finally, UK do-not-do recommendations more frequently covered mental and behavioral disorders, diseases of the genitourinary system, pregnancy, childbirth and the puerperium (see figure 1).

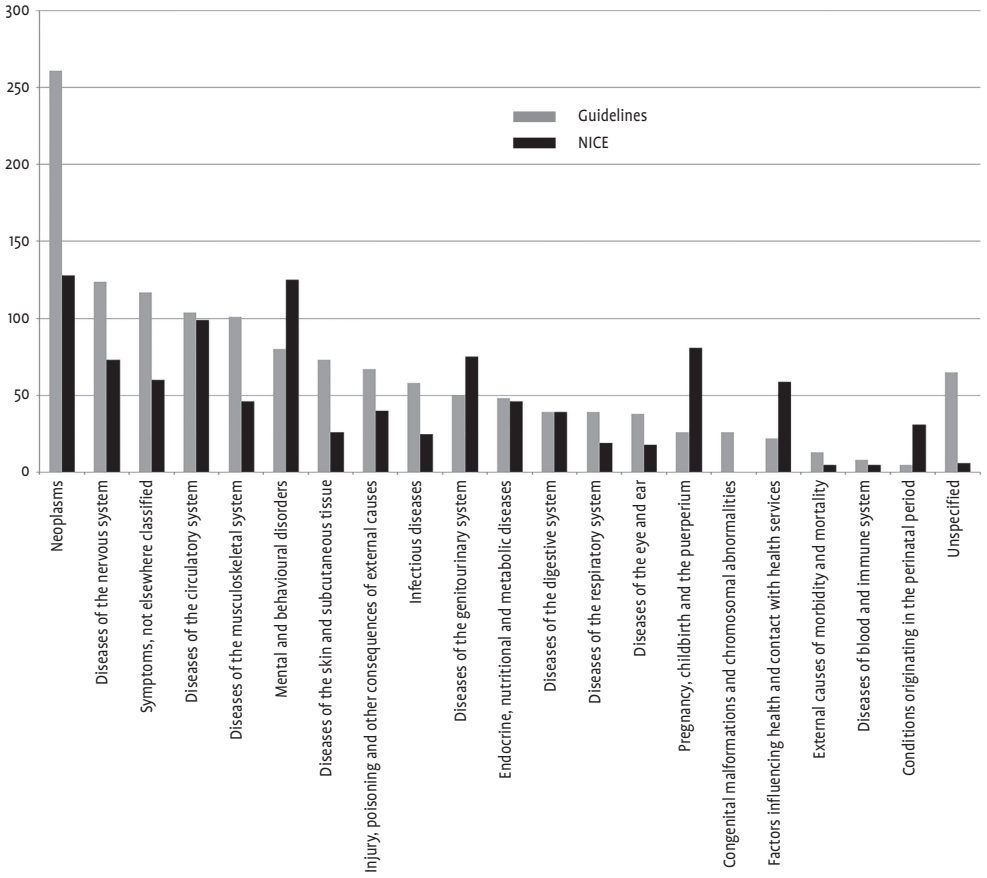


FIGURE 1 Number of lower-value services per ICD-10 group for Dutch guidelines and NICE do-not-do list.

Prioritization of Dutch lower value services

As mentioned, the ranking was performed according to two different strategies. The results of the ranking by prevalence, DALY, YLD and number of recommendations (method 1) is represented in figure 2. Both dorsalgia (back pain) and other chronic obstructive pulmonary diseases were assigned the maximum score of 16, followed by other acute ischaemic heart diseases, iron deficiency anaemia, lichen planus, and other disorders of bone (in particular the complex regional pain syndrome type 1) which each scored 14 points. Furthermore, out of the top-25 prioritized ICD10-codes, ten (40%) are in chapter M, i.e. diseases of the musculoskeletal system and connective tissue. When the ranking was performed by only prevalence and number of recommendations (method 2, figure 3), three diseases obtained the maximum score, i.e. dorsalgia, other chronic obstructive pulmonary disease, and lichen planus.

Generally speaking, neoplasm ICD10-codes receive a more modest priority when number of recommendations and prevalence are the only criteria for prioritization, but receive higher priority when burden of disease criteria are included. Ranking results for UK lower value services are provided in appendix 2.

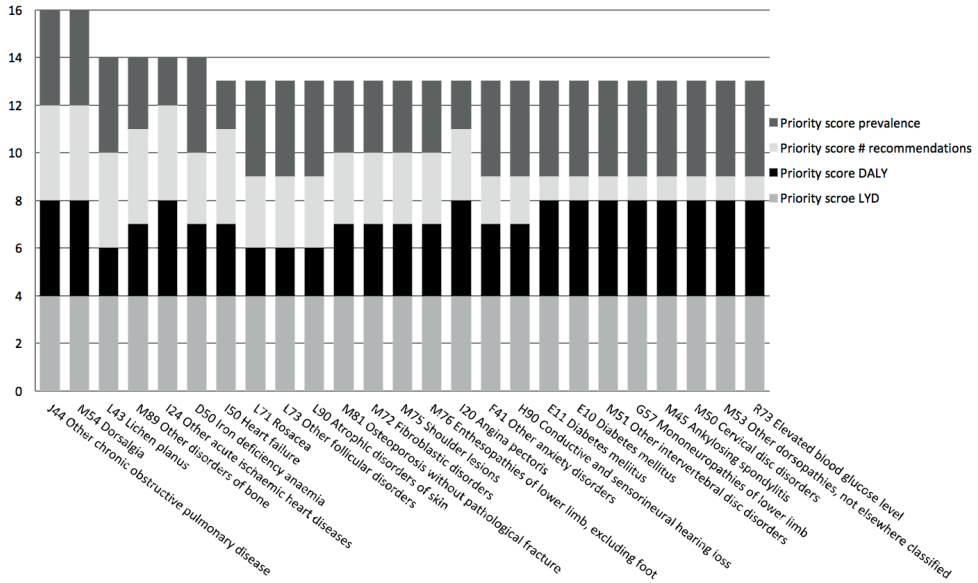


FIGURE 2 Ranking results from Dutch guidelines (method 1).

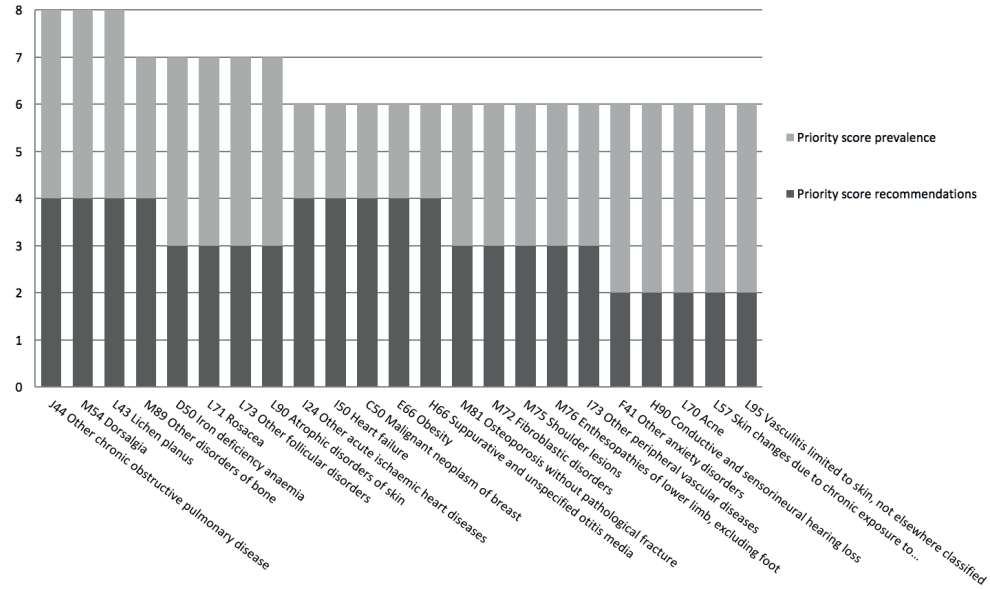


FIGURE 3 Ranking results from Dutch guidelines (method 2).

Discussion

In this study, we developed a comprehensive list of lower value services for Dutch hospital care and studied the feasibility of prioritizing the list. In addition, we repeated the descriptive analyses and prioritization for the UK do-not-do database. In total, 1366 lower value services were extracted from 193 Dutch guidelines. Of the lower value services 30% covered diagnostics, 29% related to non-drug treatment and 39% to drug treatment. The majority (77%) of all lower value services was on care that should not be offered at all, whereas the other 23% recommended on care that should not be offered routinely. ICD10-chapters that included most lower value services were neoplasms and diseases of the nervous system. Further research and policy aimed at reducing lower value services are highly warranted. A recent Dutch study showed avoidable costs are evident in healthcare: about 60 million euro can be saved in the Netherlands, when 23 lower value surgical procedures – actual use approximately 11,800 in the Netherlands – are not performed anymore [19].

The prioritization processes revealed several ICD10-codes with relatively high prevalence and disease burden where lower value services most likely occur and de-adoption is warranted, including back pain, chronic obstructive pulmonary diseases, acute ischaemic heart diseases, iron deficiency anemia, lichen planus, disorders of bone, and malignant neoplasms of bronchus and lung. These findings are relevant, given the corresponding opportunities for further research. However, this prioritization should be interpreted with caution, it does not prove lower value services are actually provided to these groups. Rather, based on robust criteria we recommend further research into the presence of lower-value services in these conditions.

The Dutch and UK list show similarities as well as differences. Dutch guidelines appear to contain more lower value services than the UK guidelines (7.1 on average vs 5.4 respectively). These data suggest Dutch guideline developers might be more aware of the existence of lower value services or might consider incorporating do-not-do recommendations in guidelines more important than their UK colleagues. However, differences in followed methodology might have spurred this difference. We only included guidelines published between 2010 and 2015, whereas NICE started in 2005, and we have shown an increase in number of do-not-do recommendations per year. Moreover, we also included recommendations from consideration-sections. This probably makes the Dutch list more comprehensive.

The development of a comprehensive list of lower value services and prioritization is only the first of several necessary steps in actually reducing lower value services, starting with measuring the actual use of lower value services. As discussed above, many uncertainties remain about the prevalence of lower value services. Estimates for the Netherlands date back to the '90s [3], or have to be gauged from case studies. Like Morgan et al [5] we support routine monitoring of potential "outbreaks" in use of diagnostics and treatment methods and variation in routine care. Such an approach entails large scale measurements using real time administrative data with sufficient clinical detail to assess appropriateness of care and risk adjustment; which are not yet available in the Netherlands. De Vries et al [27] recently identified 115 lower value care measures, which mainly focused on the cure sector. Apart from these indicators, our database could be used for developing new and valid indicators for lower value care.

Early evidence shows that dissemination of recommendations alone is not sufficient to ensure de-adoption, and that additional specific interventions are required. For example, a first evaluation of the Choosing Wisely initiative showed marginal reductions of use, if any [20], whereas Schwartz et al [30] showed alternative payment models with global budgets successfully discouraged overuse. Several papers discuss interventions or provide roadmaps for reducing overuse or promoting/advancing de-adoption [5,21,22]. Most notably, Niven et al proposed a conceptual model for the process of de-adoption; which shares much of the original Knowledge-to-Action Cycle [22,23]. The proposed framework emphasizes in-depth analyses of barriers and facilitators, which is deeply grounded in adjacent fields, such as implementation science [24]. Paprica et al [25] underlined stakeholders should be involved in de-adoption. In their analysis, they point to the trinity by Lomas et al [26] – medical effectiveness research (context-free scientific evidence), social science-oriented research (context-sensitive scientific evidence), and the expertise, views, values, and realities of stakeholders (colloquial evidence) – and show that colloquial evidence has a major influence in de-adoption. Local stakeholder involvement is therefore pivotal in de-adoption initiatives. In this study, we focused on identifying and prioritizing lower value services. This process is central to the Niven framework and is ideally performed concomitant with stakeholder engagement. Stakeholders could, for example, participate in choosing and weighting prioritization criteria. In addition, expert panels could be employed to further rank our list of lower value services on appropriateness of the services and priority for de-adoption [29].

In the Netherlands, exactly the above formula for reducing lower value care is being followed. The Dutch Federation of University Medical Centers recently initiated a four year program for reducing lower value services. The current study is the first outcome of this project and in June 2016, all eight university hospitals start local de-adoption pilot projects. The current list and prioritization contributed to selecting appropriate conditions and lower value services for de-adoption. The list will be integrated with the guideline database [31] of the Dutch Association of Medical Specialists. On this website, all lower value service recommendations will be highlighted, and special attention will be paid to the fact that in these cases not acting is a better solution.

Limitations

The methodology we developed for this study has a number of limitations, for a large part related to ambiguity in guideline recommendations and lacking data. Ambiguity in guideline recommendations sometimes made it difficult to discern lower value services, or to distinguish between care that should not be offered at all, and care that should not be offered routinely. In some cases, it was explicitly mentioned that care was not recommended, whereas in others, this was less explicit. For example, “No recommendations can be given for the use of tramadol or oxycodone in the emergency medical treatment on the basis of the emergency care literature.” (Guideline Pain management in emergency care chain). These recommendations have been included as the context shows that application is not indicated. To cope with ambiguous recommendations regular meetings were held to discuss disputable items until consensus was reached. Nevertheless, ambiguity of guideline recommendations or ambiguous populations may have biased our findings.

The Dutch list of lower value services was developed to comprehensively cover lower value services in Dutch hospital care. We restricted inclusion of guidelines to the period from 2010 until May 2015, as Dutch guidelines are recommended to be revised every five years [13]. As a result, we could not take into account important conditions or diseases covered by older guidelines, by guidelines published after May 2015 or not covered by guidelines at all. Furthermore, we might have missed some lower value services that lacked one of the keywords we identified. We therefore recommend to routinely update the list and to update the list of keywords.

Ideally, lower value services are prioritized based on the following criteria: the availability of evidence that a service is ineffective or harmful, patient safety, potential health and cost impact of de-adoption, availability of alternative practices [28] and the actual use of the lower value service. Clarifying such information for over a thousand lower value services proved impossible and much of such detailed information is currently lacking. We therefore developed alternative criteria as close as possible to the criteria proposed by Elshaug et al. Notwithstanding the methodological hurdles and data problems, we consider the prioritization results robust for singling out new and valid information besides the list itself, and both are useful for informing de-adoption programs. Finally, in this study stakeholders were not involved which should be a next step in the process of de-adoption. The prioritization results may be important input for this consultation step.

Conclusion

In this study, a comprehensive list of lower value services for Dutch hospital care was developed. The majority of lower value services covered care that should not be offered at all. Thirty percent of lower value services covered diagnostics, 29% related to non-drug treatment, and 39% to drug treatment. Comparing the list with its UK counterpart revealed that Dutch guidelines appear to contain more lower value services than the UK guidelines. Finally, a feasible method for prioritizing lower value services was established. The development of a comprehensive list of lower value services and prioritization is only the first of several necessary steps in reducing lower value services.

References

1 Institute of Medicine. *IOM definition of quality*. 2016-06-06]; Available from: http://iom.nationalacademies.org/Global/News%20Announcements/Crossing-the-Quality-Chasm-The-IOM-Health-Care-Quality-Initiative.aspx

2 Berwick, D.M. and A.D. Hackbarth, E*liminating waste in US health care*. JAMA, 2012. 307(14): p. 1513-6.

3 Grol, R. and J. Grimshaw, *From best evidence to best practice: effective implementation of change in patients' care*. Lancet, 2003. 362(9391): p. 1225-30.

4 Chassin, M.R. and R.W. Galvin, *The urgent need to improve health care quality. Institute of Medicine National Roundtable on Health Care Quality*. JAMA, 1998. 280(11): p. 1000-5.

5 Morgan, D.J., et al., *Setting a research agenda for medical overuse*. BMJ, 2015. 351: p. h4534.

6 Garner, S. and P. Littlejohns, *Disinvestment from low value clinical interventions: NICEly done?* BMJ, 2011. 343: p. d4519.

7 NICE. *Do-not-do webpage*; Available from: https://www.nice.org.uk/savingsandproductivity/collection. Accessed 06-06-2016.

8 Morden, N.E., et al., *Choosing wisely--the politics and economics of labeling low-value services*. N Engl J Med, 2014. 370(7): p. 589-92.

9 MacKean et al, *Health technology reassessment: the art of the possible*. Int J Technol Assess Health Care. 2013 Oct;29(4):418-23.

10 Prasad, V., et al., *A decade of reversal: an analysis of 146 contradicted medical practices*. Mayo Clin Proc, 2013. 88(8): p. 790-8.

11 Garner, S., et al., *Reducing ineffective practice: challenges in identifying low-value health care using Cochrane systematic reviews*. J Health Serv Res Policy, 2013. 18(1): p. 6-12.

12 Scott, I.A. and A.G. Elshaug, *Foregoing low-value care: how much evidence is needed to change beliefs?* Intern Med J, 2013. 43(2): p. 107-9.

13 Hilbing, M., M. Ouwens, and R. Kool, *De HARING-tools. Dertien instrumenten voor ondersteuning bij het opstellen, herzien, implementeren en evalueren van richtlijnen*. 2013, Scientific Institute for Quality of Healthcare, Radboud University Medical Center: Nijmegen.

14 Choudhury, M., et al. *Searching for 'do not do' recommendations from NICE guidance: a pilot study*. 2012 ; Available from: http://www.slideshare.net/HtaiBilbao/searching-for-do-not-do-recommendations-from-nice-guidance Accessed 06-06-2016.

15 Fleiss, J.L., *Measuring nominal scale agreement among many raters*. Psychological Bulletin, 1971. .76(5): p. pp.

16 WHO. *ICD-10 Version:2016*. ; Available from: http://apps.who.int/classifications/icd10/browse/2016/en Accessed 06-06-2016.

17 2013, Global Burden of Disease Collaboration. *Global Burden of Disease Study 2013* (GBD 2013) Data Downloads. ; Available from: http://ghdx.healthdata.org/global-burden-disease-study-2013-gbd-2013-data-downloads. Accessed 06-06-2016.

18 Landis, J.R. and G.G. Koch, *The measurement of observer agreement for categorical data*. Biometrics, 1977. 33(1): p. 159-74.

19 Govers, T.M., et al., *Lower value surgical procedures create a significant waste in healthcare.* Submitted, 2016.
20 Rosenberg, A., et al., *Early Trends Among Seven Recommendations From the Choosing Wisely Campaign*. JAMA Intern Med, 2015. 175(12): p. 1913-20.
21 Colla, C.H., *Swimming against the current--what might work to reduce low-value care?* N Engl J Med, 2014. 371(14): p. 1280-3.
22 Niven, D.J., et al., *Towards understanding the de-adoption of low-value clinical practices: a scoping review*. BMC Med, 2015. 13: p. 255.
23 Graham, I.D., et al., *Lost in knowledge translation: time for a map?* J Contin Educ Health Prof, 2006. 26(1): p. 13-24.
24 Grol, R. and M. Wensing, *Implementatie. Effectieve verbetering van de patientenzorg*. Vol. 4. 2011.
25 Paprica, P.A., et al., *From Talk to Action: Policy Stakeholders, Appropriateness, and Selective Disinvestment*. Int J Technol Assess Health Care, 2015. 31(4): p. 236-40.
26 Lomas, J., et al., *Conceptualizing and combining evidence for health system guidance*. 2005, Canadian Health Services Research Foundation: Ottowa, Canada.
27. de Vries, E. F., et al. Are low-value care measures up to the task? A systematic review of the literature. BMC Health Serv Res 2016(1): 405.
28 Elshaug, A, G, et ak, Identifying existing health care services that do not provide value for money. Med J Aust. 2009 Mar 2;190(5):269-73.
29 Scott, I. A. and S. J. Duckett (2015). "In search of professional consensus in defining and reducing low-value care." Med J Aust 203(4): 179-181.
30 Schwartz, A. L., et al. (2015). "Changes in Low-Value Services in Year 1 of the Medicare Pioneer Accountable Care Organization Program." JAMA Intern Med 175(11): 1815-1825.
31 www.richtlijnendatabase.nl

Appendix 1. prioritization methodology

4a. Connection with Global Burden of Disease

Since information on prevalence and disease burden by ICD10-code is not systematically available in the Netherlands, it was decided to use the Global Burden of Disease (GBD) classification. The GBD is a global classification of disease categories including prevalence, disability adjusted life years (DALYs), years of life lost (YLL) and years lost due to disability (YLD) for each disease category, and is regularly renewed, most recently in 2015[1] with 2013 data. Many of the GBD parameters are available online and by country. Also, information was available which ICD10-codes are covered by each GBD-category. A GBD-category often covers a range of ICD10-codes. For example, the GBD-category "low back pain" consists of a number of ICD10-codes from Chapter VI – Diseases of the nervous system (including disorders of lumbosacral plexus), but also from Chapter XIII – Illnesses of the musculoskeletal system and connective tissue (including instability of spine).

The ICD10-codes were assigned to GBD-categories. If an ICD10-code fitted in more than one GBD category, the category covering the smallest range of ICD10-codes was chosen. Part of the ICD10-codes only fitted in the 'garbage code' category, which is a very broad group. Therefore, for those ICD-10 codes that were assigned to the rest category 'garbage code', an alternative GBD-category was sought, if possible. If no ICD10-code was assigned to a lower value service but an ICD10-group or specialism was known, this information was used to find an appropriate GBD-category for the lower value service. Four researchers (MEAM, EV, JW, TA) each took a randomly selected part of the list to assign GBD-categories to ICD10-codes. Equivocal cases were discussed with another researcher until consensus was reached. Not for each GBD-category prevalence figures were known, and to a lesser extent, DALYs and YLDs were missing.

4b. Prioritization of ICD10-codes

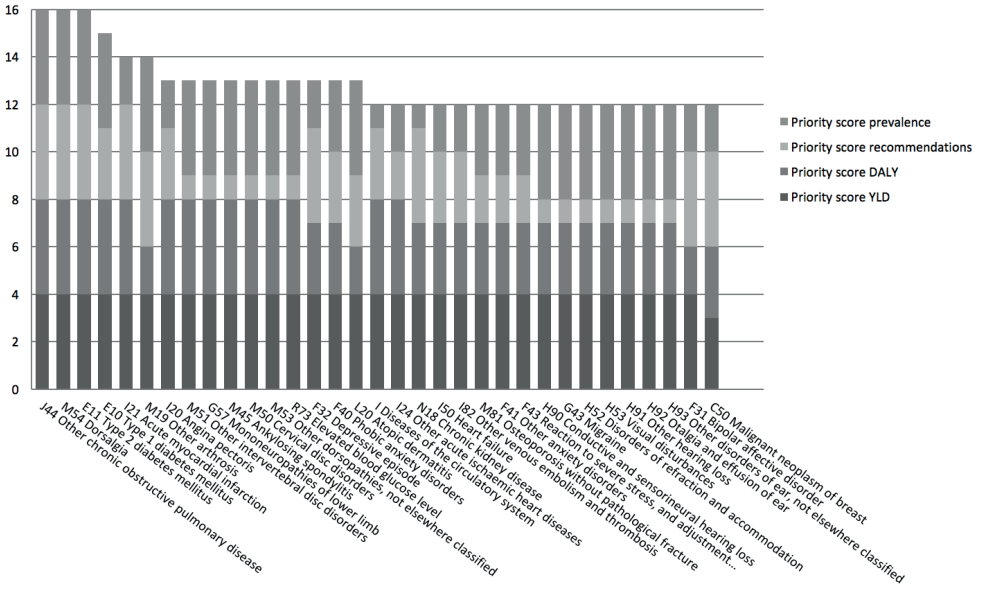
To apply a prioritization in the extensive list of lower value services, the following criteria were defined per ICD code: number of lower value services, prevalence, LYD and DALYs. For each criterion four groups were made that were given a priority score. The classification into four groups aimed at having a comparable number of ICD10-codes in each group, as well as obtaining rounded categories (i.e. 500-1000 instead of 439-768). This resulted in the following classification:

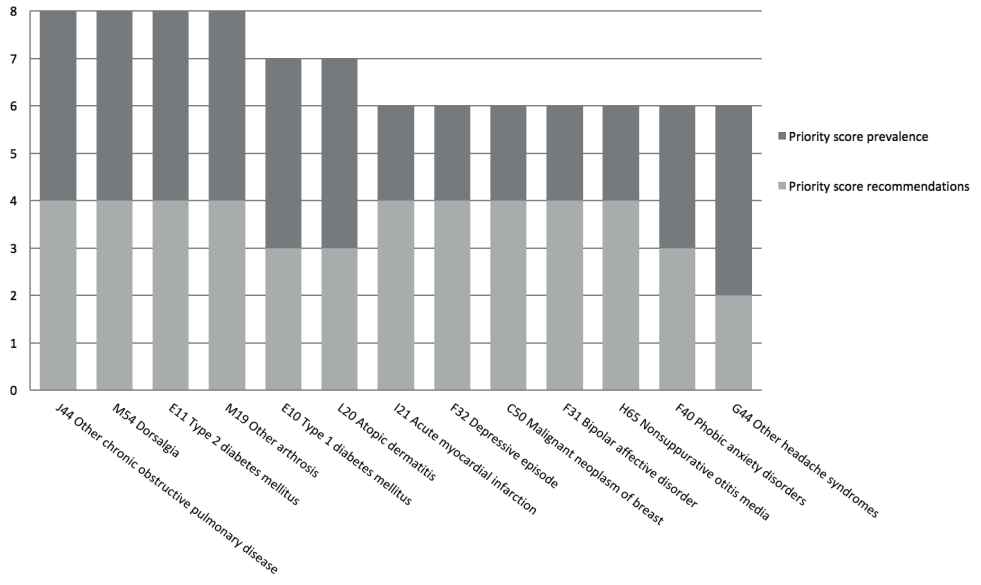
- Number of lower value services per ICD10-code: >10 (4 points), 5-10 (3 points), 2-5 (2 points), 1 (1 point);
- Prevalence (Netherlands 2013;*1000): >1000 (4 points), 500-1000 (3 points), 100-500 (2 points) , <= 100 (1 point);
- YLD (Netherlands 2013;*1000): >10 (4 points), 5-10 (3 points), 1-5 (2 points), <=1 (1 point);
- DALY (Netherlands 2013;*1000): >100 (4 points), 50-100 (3 points), 10-50 (2 points), <=10 (1 point).

1 Global Burden of disease Study Collaborators 2013; http://ghdx.healthdata.org/global-burden-disease-study-2013-gbd-2013-data-downloads Global Burden of Disease Study 2013. Global Burden of Disease Study 2013 (GBD 2013) Incidence, Prevalence, and Years Lived with Disability 1990-2013. Seattle, United States: Institute for Health Metrics and Evaluation (IHME), 2015.

Subsequently, the ICD10-codes were prioritized by the sum of the number of lower value services, prevalence, LYD and DALY (Method 1), the highest score (up to 16) indicating the highest priority. As YLD is part of the DALY, this provides a high priority for ICD10-codes with a high number of YLD. Therefore, the prioritization was repeated for the sum of the number of lower value services and DALYs (Method 2; maximum score 8).

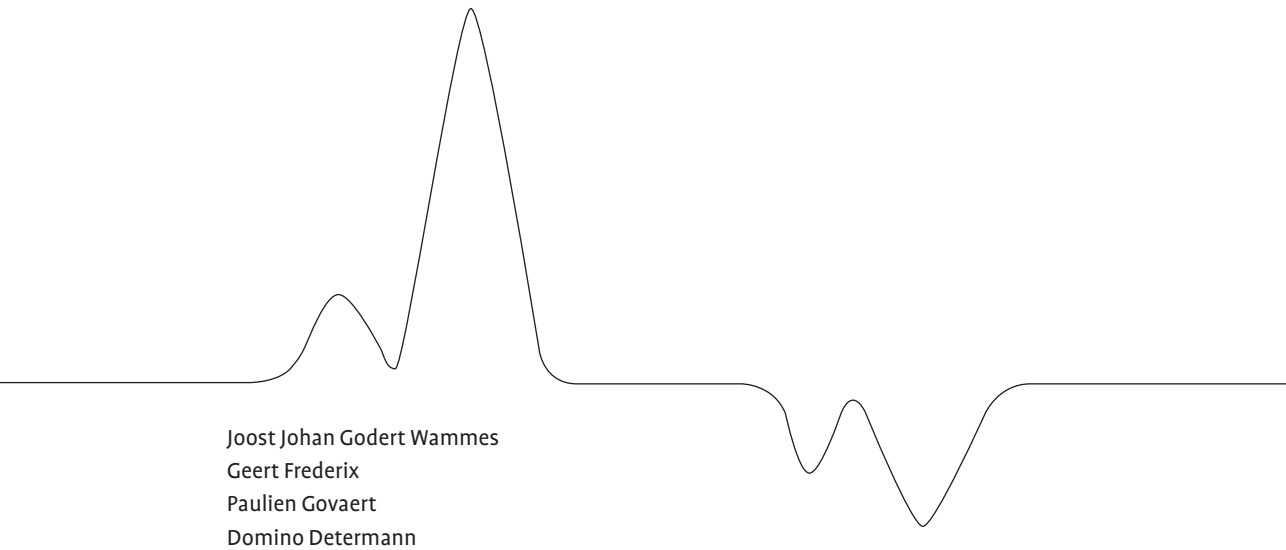
Appendix 2: UK prioritization results





CHAPTER 5

Displacement effects in Dutch hospital care: a managed competition setting



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Abstract

Under the circumstance of a constrained budget, cost-increasing technologies may displace funds from existing health services. It is highly uncertain however, what services are displaced and how such displacement takes place in practice. In the current research, we understood displacement as a process, a total of decisions made after the introduction of a new cost-increasing technology; a pathway from new technology to opportunity costs. We conducted six case-studies and interviewed 84 professionals with various roles and responsibilities (practitioners, departments, board of directors, insurers, and others) to investigate how Dutch hospitals have dealt with the cost pressure of cost-increasing health technologies. Transcripts were analyzed thematically in Atlas.ti on the basis of an item list.

Our findings show that the opportunity costs of cost-increasing health technologies are not easily identifiable; limited transparency in the internal allocation of funds within a hospital contributed to this. Furthermore, we found that the entry of innovations/new technologies and cost-containment are two parallel processes that are generally not causally linked. The way of financing is pivotal in displacement in the Netherlands, because there is a separate budget for expensive drugs. This budget pressure is reallocated horizontally across departments, whereas the budget pressure of remaining services is primarily reallocated vertically within departments or divisions. Hospitals have reacted to budget pressures primarily through a narrowing in the portfolio of their services, and a range of (other) efficiency measures. The board of directors is central in these processes, insurers are involved only to a limited extent. Direct displacement of high-value care due to the introduction of new innovations was rarely observed. Rationing (primarily reducing accessibility) was observed mainly in response to cumulative cost pressures, production ceilings and capacity problems. Active surveillance of waiting lists is warranted to prevent waiting list driven morbidity.

Introduction

In the Netherlands a broad agreement between stakeholders in the healthcare sector has been agreed on, among other things, maximum permitted budgetary growth (1.3% in 2019, decreasing to 0% in 2022, excluding wage and price adjustment). Budget pressure is further increased by the continuous introduction of cost-increasing health technologies. Decision makers, both at local and national levels, therefore have to make choices about how to spend their resources. At the national level, the Health Care Institute (ZINL) of the Netherlands advises the Minister of Health (MOH) on the contents of the basic benefit package. In 2016, ZINL advised the MOH not to reimburse two expensive drugs – Pertuzumab (Perjeta®) and Pembrolizumab (Keytruda®) – unless their cost-effectiveness would be improved and budget impact would be less through price negotiations. The main argument was that, in current economic circumstances, reimbursement of these drugs could require displacement of more cost-effective services, resulting in a net loss of health benefits at the population level [1,2]. Based on this advice, the Minister negotiated lower prices with the manufacturers and decided that the drugs would qualify for reimbursement until the end of 2019 [3]. These negotiated prices are not revealed in the public domain.

In England, Wales and Scotland, research into displacement has mainly focused on estimating the cost per QALY threshold [4,5]. Little is known however, about displacement in practice, how displacement takes place and what services are displaced to accommodate new cost-increasing technologies. We know of only one study of the introduction of cost-increasing technologies. This Welsh study investigated how NHS commissioners accommodated financial 'shocks' originating from Technology Appraisals issued by NICE. They found that the 'displacement assumption' (existing services are displaced to accommodate cost-increasing technologies) generally did not hold; and that financial shocks originating from Technology Appraisals were generally accommodated by greater efficiency and increased spending. In addition, commissioners sought for savings or efficiency measures in response to cumulative cost pressures from multiple sources rather than in response to single Technology Appraisals [6]. One limitation of this research was that it was limited to decision making of Finance Directors and Medical Directors of Local Health Boards in Wales. Although directors may have the best oversight of macro and meso level decision making, they may have been unaware of decisions made at lower organisational levels in their geographical areas. Besides, their findings may not be representative for countries with other organisational and financial structures.

In the current research, we understood displacement as a process, a total of decisions and resulting consequences made after the introduction of a cost-increasing technology. This includes priority setting at higher organisational levels and bedside rationing at lower organisational levels. Insight in displacement also requires exploration of the causality of resource allocation ((how) does one decision leads to another?) and the ultimate effects for individual patients. We defined priority setting as resource allocation decisions between different services, patient groups, or elements of care; whereas bedside rationing was interpreted as the effects of such decisions on individual patients [7]. Priority setting in general has been researched for many years, albeit relatively little attention has been paid to the impact on individual patients.

The aim of our study was to investigate how Dutch hospitals have dealt with the introduction of cost-increasing health technologies, and to investigate the link between the inflow of new technologies and outflow of existing technologies. Specifically, we aimed 1) to test whether displacement takes place; 2) to identify which services, patient groups or elements of care are typically displaced; and 3) to identify the main actors that prioritize and ration and to investigate the motives for such decision making. Six case-studies were conducted to understand similarities and differences between cases, and to investigate the mechanisms of displacement and how these relate to financial and organisational structure.

Institutional background

In the Netherlands, nearly universal coverage for curative care is achieved through mandatory purchase of statutory health insurance from private insurers. The Health Insurance Act legally requires health insurers to provide a comprehensive nationally set benefits package. Decisions regarding the package rest with the Minister of Health, who relies on advice from the National Health Care Institute and its Healthcare Insurance Board [8,9]. Coverage of prescription drugs is described in positive lists. Remaining service coverage is specified through an open specification with a general (functional) description of benefits, and restrictions are expressed in negative lists [10]. The great majority of services enter the health system without formal assessment through this 'open' specification.

The Healthcare Insurance Board uses four criteria to determine whether or not to reimburse a new health service: necessity, effectiveness, cost-effectiveness, and feasibility for implementation. Adoption of a technology is based on an integral assessment on the basis of these criteria, the criteria are not used as knock-out criteria. The criteria are continuously refined and improved, and especially the cost-effectiveness criterion is debated. In 2006, the RVZ (government advisory body) argued that treatments with a cost-effectiveness ratio higher than €80.000/QALY should not be included in the basic benefit package. The RVZ also stated that the acceptable costs per QALY vary according to burden of disease and other factors, such as rarity of the disease [11]. In reality however, treatments are rarely excluded from the basic package based on 'unacceptable' cost-effectiveness. In addition, besides the appraisal criteria several other factors have played a role in defining the actual constituents of the basic benefit package, including the desire to control costs and the public opinion [12].

The Dutch healthcare system is largely based on the principles of managed competition with little central planning. Health care purchasing is considered the centrepiece of the system and is the main instrument for stimulating efficiency. Insurers are supposed to prudently purchase care for their enrolees. In real life, insurers and hospitals mainly negotiate on volumes and prices, while quality of care plays only a minor role in these negotiations. In addition, insurers may decide not to contract a provider (selective contracting), but are required to offer adequate care for their enrolees. The great majority of hospital care in the Netherlands is reimbursed through payment products similar to Diagnosis Related Group (DRGs; which cover both in- and outpatient hospital care). About 70% of DRG-prices are freely negotiable, the rest of the prices are regulated. A small part of hospital care is reimbursed through so-called add-ons. Add-ons are separate payments that have been developed for the reimbursement of expensive drugs and intensive care unit admissions.

From 2012 onwards, the Minister of Health has made sector agreements with providers and insurers that have effectively limited spending growth to 2.5% during 2012 and 2013, 1.5% in 2014 and 1% in 2015-2017. Insurers and hospitals negotiate prices and volumes on a yearly basis, guided by the terms of the sector agreements. Because of the sector agreements, it has been argued that hospitals and insurers de facto negotiate lump sum contracts with revenue ceilings as the most important provision. In addition to the ex ante contracts with stipulated prices and volumes, a small part of hospital spending – for 'non-steerable' and very expensive services, including transplant care and expensive drugs – is carved out from the revenue ceiling and funded on a fee for service basis.

Materials and methods

Study design

We chose a multiple qualitative case study design to study displacement in the hospital sector of the Netherlands. Case studies are well suited to explore, deconstruct and reconstruct social phenomena, which we expected the displacement process to be. The design is based on a constructivist paradigm that is built upon the premise of a social construction of reality [13]. Our aim was to obtain the experiences and perspectives of a diverse range of stakeholders that have been involved in displacement decision making processes. We conducted six case studies to be able to understand similarities and differences between cases. Prior to this work, a pilot study was conducted to inform and to test the feasibility of our approach. Halfway through the project we organized an expert meeting with national experts (N=9) in health economics and policy to discuss preliminary findings.

We purposefully chose six cost-increasing health technologies. First, a stakeholder meeting with our funder (ZINL) was held to identify case studies meeting a pre-specified set of criteria. In addition, we searched several (government) websites and explored cases through our personal networks. Apart from maximum variation, interventions were required to meet the following criteria: 1) interventions should be generally considered cost-ineffective based on current Dutch standards 2) interventions should be provided in hospitals 3) the reimbursement decisions should have been made some time ago, in order to be able to identify possible displacement effects 4) the intervention should have a relatively high budget impact. Based on these criteria, we chose intramural oncolytics, robotic (Da Vinci) surgery, Left Ventricular Assist Device, endovascular aneurysm repair, population screening for colon cancer, and expensive eye injections (Eylea and Lucentis). A short description of the interventions is given in table 1.

Participant selection and recruitment

We purposefully selected key stakeholders (experts for the particular health technology) to be interviewed for the case studies. Key stakeholders were initially identified from policy documents, websites, the media or from our (funder's) network. We then asked the initial key informants to suggest other participants (snowball sampling). We aimed to take into account geographic spread, to recruit a diverse set of relevant stakeholders, with different positions and responsibilities, per case study as well as per hospital within a case study.

TABLE 1 Description of the case studies.

Innovation	Description
Left ventricular assist device (LVAD)	LVADs are devices for assisting cardiac circulation. They have been used from 1992 onwards as a 'bridge to transplant' for patients with advanced heart failure. During the years, the outcomes of the therapy have steadily improved, such that LVAD can be used as long term therapy ('destination', LVAD is not followed up by a heart transplant).
Fenestrated endovascular aneurysm repair	In this procedure an expandable stent graft is placed within the aorta to treat aortic disease. This minimally-invasive technique is indicated for high-risk patients unfit for open surgery. Fenestrated and branched EVAR (FEVAR) are expensive due to its custom-made graft device.
Expensive oncolytics	In recent years, several relatively expensive oncolytics have been approved for inclusion in the basic benefit package, including pertuzumab, palbociclib, nivolumab, pembrolizumab, atezolizumab, and ibrutinib.
Eylea and Lucentis	Avastin, Eylea and Lucentis are all used for the treatment of various eye diseases. Eylea and Lucentis are both much more expensive than Avastin, but are equally effective for most indications. Eylea and Lucentis are indicated for patients for whom Avastin is not effective, and for patients with diabetic macula oedema and vascular occlusion.
Population screening for colon cancer	In 2014, the Netherlands started population screening for colon cancer. People with positive test results are advised to get a colonoscopy in the hospital. Studies have shown that this surveillance is not cost-ineffective [14].
Robotic surgery	Robotic assisted minimally invasive surgery has been performed in the Netherlands since 2000, as an alternative to 'pure' laparoscopy or open procedures for various indications. Despite many studies, there is still no clear-cut evidence regarding the cost-effectiveness.

For example, we aimed to recruit informants from general, specialized as well as academic hospitals, and we recruited medical doctors, financial managers, sales managers, board members, and health care purchasers (insurers). Participants were invited to participate in the study by e-mail. The invitation letter provided a summary of the aim and methodology of the study, as well as the time needed for the interview. We sent reminders when we did not receive a response within two weeks.

Data collection

The interviews took place between September 2016 and May 2017. All interviews per case study were conducted by one single interviewer. The interviews and analysis were undertaken concurrently and iteratively, in order to inform subsequent interviews. The interview team met at least monthly during the duration of the study to discuss the findings and to coordinate ongoing work.

The primary aims of the interviews were to identify the main (financial) consequences of the introduction of the particular health technology for the department, hospital or

insurer; and to discuss the choices and decisions that were made, as well as the reasons for making the decisions. We also more generally discussed displacement processes and the mechanisms known to the respondent that were beyond the case study.

A semi-structured topic guide was used for the interviews, including the introduction process of the health technology; agreements and negotiation processes with third party payers; problems encountered (costs, time, facilities, etc) due to the introduction of the health technology; what action was taken in response to the problems; the consequences for care provision and rationing; and views concerning displacement (the interview guide is presented in appendix 1). Rationing was operationalized according to Klein's rationing strategies, including rationing by denial, selection, delay, deterrence, deflection and dilution [7].

The topic guide was based on relevant literature and a pilot study, and adapted based on the first five interviews. We made minor amendments to the topic guide for our interviews with hospital boards members and health insurers. During these interviews we discussed multiple case studies, and more time was devoted to displacement processes and priority setting in general.

The interviews were predominantly conducted face to face, at the respondent's office, but twenty interviews were conducted by telephone. Interviews ranged 20-60 minutes and were audio-recorded and transcribed verbatim. Permission for audio recording was sought for and given in every case. The purpose of the interview and the general aim of the study were summarized at the start of each interview. We explained that neither findings nor quotes would be attributed to individuals or organizations.

Data analysis and presentation

Transcripts were transcribed verbatim and analyzed thematically in Atlas.ti on the basis of an item list. This item list was derived from the semi-structured topic guide and literature. We made minor modifications to the topic list based on discussions within the broader study team. All analyses were performed by the interviewer who held the interview. In case of data ambiguity we contacted the respondents to retrieve the meaning of a quote. For the purpose of inter-researcher reliability, the interviewers met regularly to discuss themes and data categories. In addition, at the start of the analysis, at least one transcript per case study was independently coded by two or more researchers and the results were compared. Any differences in data interpretation were discussed and resolved.

We developed summary tables of the case studies according to the categories of the item list and compared the results across the case studies to identify systematic patterns of displacement. Based on this information, a narrative summary of the results was made. The results are presented according to the flow chart below (figure 1) which follows the budgetary flow in the health system and our interview guide. The arrows of the flowchart indicate how stakeholders can (re-)allocate the budget pressure, either upwards (left side), or downwards (right side). The green circles correspond with the paragraphs in the results section.

We first present respondent characteristics, and then discuss the introduction process of the health technology and actors involved; agreements and negotiations with insurers; problems encountered (costs, time, facilities, etc) due to the health technology; decision making process in response to the problems; the consequences for care provision and rationing; and views concerning displacement.

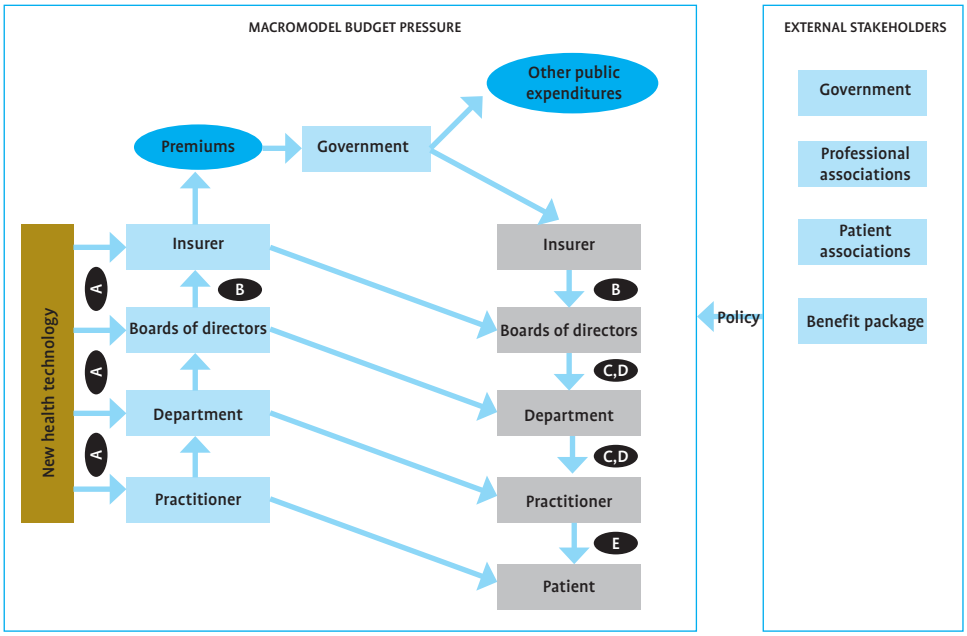


FIGURE 1 Macro model budget pressure, stakeholder model.

TABLE 2 Interview informants and roles per case study.[1]

	Left ventricular assist device	Fenestrated endovascular aneurysm repair	Expensive oncolytics[2]	Eylea and Lucentis	Population screening for colon cancer	Robotic surgery
Medical doctors	5	8	12	8	8	6
Managers/directors	2	3	1	1	2	2
Professional scientific associations			1	1	1	1
Patient associations			1	1	1	1
Manager sales	1	3	1			1
Board of directors	1	1	5	4	4	2
Insurer		1	1	1	1	2
Hospital pharmacists			3			
Other			1		1	

[1] Informants may be listed on several roles or columns. For example, a medical doctor may be a part-time member of the sales team of the hospital, or a member of the board of directors may have spoken about two or more case studies.

[2] Including the pilot study.

Results

Respondents

In total 84 interviews were conducted. Table 2 presents the characteristics of the respondents for each of the case studies. A minimum of nine interviews were held per case-study. Medical doctors were generally overrepresented among our respondents. However, in each of our case studies we interviewed a diverse range of stakeholders, with at least four distinct roles and responsibilities in the Dutch health system.

Below the main findings of the interviews are presented. Paragraph A and B provide main contextual findings concerning the health technologies; which are essential for understanding the displacement mechanisms that will be outlined in paragraph C until E.

A Entry to the system and hospital

An elaborate description of our research findings per case study is presented in appendix 2. All but one of the technologies have been assessed by ZINL for inclusion in the benefit package. LVAD-destination and FEVAR were initially not included in the benefit package, but were adopted later when new scientific evidence concerning the benefits of the treatments became available. ZINL advised the government not to include Lucentis in the benefit package, but this advice was not acted upon. The Da Vinci platform was assessed, but not formally in- or excluded, because decisions about service coverage concern treatments for a given patient population, not the way the treatment is delivered.

The entry into hospitals also differed between the cases. In most of the case studies, a wide range of stakeholders were involved in decision making processes, including health professionals, managers of hospital departments, board of directors, investment or drug committees, and in some cases also stakeholders from outside the hospital (medical societies, healthcare inspectorate, ZINL, other governmental agencies). However, in case of FEVAR, specialists and departments started experimenting with one or a few test procedures, before activities were scaled up after which stakeholders at a higher hierarchy level of the hospital were involved. LVAD, FEVAR, Lucentis and Eylea were all used at considerable scale before they were formally included in the basic benefit package.

B Reimbursement, contracts and negotiation with insurers

In the Netherlands, hospitals and insurers negotiate 1) carved out contracts for expensive services, using add-on payments based on fee for service and without cap (in Dutch “nacalculatie”) and 2) ex ante a revenue ceiling contract based on prices and volumes (in Dutch “plafondafspraken”). Although the agreements allow for differentiation in percentages growth per hospital, the growth norm is used as a guiding principle for the negotiations. During the year, hospitals and insurers discuss new interventions and policy on a continuous basis. In autumn, new contracts for the upcoming year(s) are negotiated.

The carved out contracts primarily include expensive drugs and expensive procedures such as LVADs. The budget for expensive drugs is not part of department budgets, but is a separate budget. Hospitals and insurers negotiate the volume and price of expensive oncolytics, and sometimes also an ex ante determined capped budget. Generally speaking,

the reimbursement of oncolytics is undisputed, and insurers will reimburse on basis of fee for service, as they fear for loss of reputation. However, indication extension during the year has led to budget overruns in some hospitals that were unable to negotiate extra money. There are several requirements for carved out contracts, including guideline adherence, transparency, and no margins on the drugs.

Negotiations about the ceiled revenue are parallel to, or subsequent to negotiations of the carved out contracts. Hospitals generally prepare long lists of investment opportunities, and similarly insurers prepare lists of disinvestment opportunities. Occasionally individual items of such lists are discussed and accepted or rejected. However, generally speaking, hospital and insurer primarily negotiate a revenue ceiling, which is secondarily based on prices and volumes. Terms about specific services are not binding, and may be exchanged for any other services. Cross-subsidization (services are paid from the margins of other services) was widely reported.

C Problems encountered

The interviewees reported a wide range of problems they were faced with when the intervention was introduced. In case of LVAD and population screening for colon cancer, participants reported predominantly capacity problems (increased need for specialized personnel, operating room capacity, intensive care beds) and only limited financial problems. Below the most important financial problems, and problems intrinsically related to displacement, are outlined.

C1 Investment opportunities exceed the permitted growth

Both insurers and hospital management generally did not doubt the added value of most investment opportunities. However, it was clear that the associated total costs could not be accommodated in the current growth path. Many respondents argued that the increase in expensive drugs was at the expense of other services. It was hard to say however, at what expense exactly.

> *"If that were not the case, then the rest of the negotiations might have been a lot easier. The expensive drugs are the elephant in the room."*
> BOARD OF DIRECTORS

Respondents generally pointed to the totality of budget increases, rather than to the growth of individual drugs or services. For example, FEVAR was one of a range of services contributing to the cumulative budget overrun. As a result, what could be observed is a competition between technologies and services for spending growth.

> *"Instead, we do complicated things, like FEVAR-prostheses, complicated laparoscopic operations and so on. That costs twice as much, but our budget does not grow. So at the meso level of the department, there is a continuous fight with the Board of Directors."*
> VASCULAR SURGEON

From the interviews it appeared that the degree of experienced cost pressure differed between settings, depending on the financial organisation of the hospital and negotiating power. Generally speaking, we found that the cost pressure in surgical departments/divisions was more severe than in cardiology/cardiothoracic divisions. In addition, respondents argued that smaller hospitals face higher risks for cost pressure due to expensive drugs, as they were less likely to negotiate generous contracts with insurance companies. In the eye drugs case, specialized eye centres experienced heavier cost pressure, as they had less abilities for cross-subsidization or abilities to exchange services.

C2 The distribution of flow of funds within a hospital is not transparent

Especially in larger and academic hospitals, many revenue sources exist, including innovations funds, education fees, research funds and others. Consequently, hospitals use internal funds in which the various revenue sources are reallocated (services were exchanged, or through cross-subsidization).

> *"We work with a budget system. We negotiate about how that budget is built, but it is up to the healthcare provider how to fill in that budget. A healthcare provider always has the possibility to reallocate the money somewhere else instead of to that DRG."*
>
> INSURER

Respondents reported a lack of transparency in the hospital's internal financing. In the current system, negotiated DRG-prices may not represent real prices, and hospitals may lack insight in the costs of their DRGs. Negotiations rarely take place on intervention or technology level and are mostly based on hospital revenue deals. Consequently, the additional costs of an intervention or displacement effects are hardly visible.

> *"The system is not so one-dimensional that such effects are immediately visible and you get a difficult conversation about the disposables. There are many possibilities and sources for substitution."*
>
> SURGEON

There was a lot of unawareness about negotiations with insurers. Most respondents named volumes for services, which were also used for internal planning (they were not necessarily contracted). In some hospitals, managers at higher levels were unaware of individual prices (or profit margins) for services in their budget, or accepted business-cases.

D Decision making processes, underlying reasons and contra-mechanisms to budgetary pressure

D1 Decision making differs across types of financing, board of directors are central to decision making

Priority setting and rationing within hospitals differs depending on the type of service and type of budget. Expensive drugs requests are assessed by drug committees, before the Board

of Directors are involved, who may negotiate additional budget from insurers. Because there is a separate budget for expensive drugs, the budget pressure is experienced at the higher managerial levels, not only by the department that uses the drugs. This budget pressure is accommodated by insurers, and indirectly by departments in the hospital through lower department budgets.

In addition to the budget for expensive drugs, the hospital budget is cut into budgets for divisions and (sub-)department. Departments and divisions are relatively free in how to spend this budget, but they are kept relatively strictly to this budget. They discuss their policy, budgets and activities with the board of directors on a regular basis.

> *"Look, if the cardiologists want to grow in the field of interventions, then maybe they should not grow in the area of the fast-track outpatient clinics. "*
> BOARD OF DIRECTORS

Departments and divisions may submit business-cases to request additional funding. The board of directors (and sales team) are central to this decision making. They may decide to include the business-case in negotiations with insurers (external business-cases). In exceptional cases (long waiting lists) hospitals have successfully negotiated extra funding. Internal business-cases are not discussed with insurers and may be rejected, or funded from other sources. Most of the times, the board will request the departments to take austerity measures (see paragraph B). In each of the cases, the board of directors were involved in introductory decision making or growth.

D2 Strategic considerations and key topics

A range of arguments were mentioned for introducing a technology, or to further invest in the growth of a particular service. In all case studies, patients were expected to benefit from the treatment. In addition, respondents argued that the technology was considered a key topic of the department and the hospital. Such emphasis on key topics can take many forms, and key topics were chosen at every managerial level (e.g. from high to low: cardiovascular centres, vascular surgery, aorta pathology). Generally speaking, such key topics receive more funding, at the expense of others.

> *"In the coming years in particular that cardiothoracic and vascular domain will grow, maybe at the expense of others. That we say in other respects, that is no longer for us."*
> BOARD OF DIRECTORS

Besides, several respondents pointed to competition between providers: providers were afraid to lose patients, or were afraid to stay behind technologically.

> *"If we limit that flow of patients, then we will lose it, then they'll look for someone else. Until today, that was one of the reasons why we accept the budget overrun."*
> SURGEON

Some respondents were critical about the decision making, doubted the benefits of the treatment and noted that a clear evidence base was lacking. Cost-effectiveness was rarely considered. Besides, respondents argued the industry had 'pushed' the innovation too much.

D3 Contra-mechanisms

We asked the respondents how they dealt with the budget pressure of the new technologies. Respondents primarily pointed to their choices in the portfolio of their services. Insurers and the board of directors request departments to stop providing services that can be provided elsewhere at lower costs.

> *"Someone with a minor heart attack, and when treatment has gone straightforward, should simply be followed-up elsewhere."*
> CARDIOLOGIST

Some respondents doubted the budget impact of such measures, albeit it effectively reduced work load. Hospitals increasingly collaborate in this re-arrangement of service delivery, but the degree of collaboration differs considerably across hospitals.

A variety of other measures to relieve budget pressure or capacity problems were mentioned, including effort to reduce the price of LVADs, FEVAR-stents, and expensive drugs. Many doctors stated that they adhered to guideline recommendations more strictly than before, or that eligibility criteria for procedures or drugs were redefined. Besides, efforts were taken to reduce the length of stay or to technically improve services. Task rearrangements, substitution, e-health, and cuts in staff and beds were also mentioned.

E Displacement, and impact on regular care

We asked interviewees directly which services were displaced to accommodate the introduction of the innovation, and which effects this had for regular care and for individual patients. In case of LVAD, respondents pointed to generous financing, and that problems primarily occurred due to capacity constraints. FEVAR was one of the services contributing to cumulative cost pressures, and in some hospitals FEVAR was rationed due to cost pressures from other services. The budget pressure of expensive drugs was accommodated by insurers and the board of directors, who redistributed this to the rest of the departments (horizontal reallocation). In the eye drugs case, rationing was widely reported, but cost pressure was only one of the several factors that necessitated rationing. Population screening for colon cancer was also rationed, but this was primarily due to shortages in GE-specialists. The additional costs for Da Vinci surgery were largely unknown, and cross-subsidized from other services.

With few exceptions, there was consensus that displacement, and efficiency/austerity measures were not causally linked to investments in technologies.

> *"I cannot but remember that we had to cut costs and look for efficiency gains. But I cannot say that this really is at the expense or coincides with that Da Vinci. That is a permanent system to level the costs and the revenues."*
> MANAGER UROLOGY

Although not necessarily related to the technologies, many respondents pointed to current pressures in Dutch hospitals, and the necessity to ration care. Below the most important mechanisms are outlined.

E1 Rationing is usually the result of production ceilings and capacity problems

Many respondents noted that rationing was the result of cumulative pressure from several sources, including aging, reform in long term care, and technological innovation. Shortages in personnel and beds further complicated the situation. Occasionally, but not structurally, such capacity problems were related to austerity measures. Furthermore, individual services were rarely rationed, but rationing occurred rather in larger organizational units, such as surgical divisions, or cardiovascular centres. Several respondents blamed the sector agreements and argued that insurers do not purchase enough care.

E2 Hospitals primarily reduce accessibility in response to cost pressure

Respondents listed all rationing strategies; and rationing by delay was mentioned most frequently and was regarded the primary rationing strategy. In case of a budget overrun or capacity problems, the board of directors request departments to reduce accessibility.

> *"Yes, then we consult with the manager and the head of the department, and tell them to increase the waiting lists."*
> BOARD OF DIRECTORS

Rationing strategies were usually combined, especially rationing by delay and selection were often used in tandem. Furthermore, patients were prioritized on the basis of medical need: malign and acute patients got direct access, while benign and non-acute patients were queued.

> *"If your operation room time is limited and you have to choose, the oncology patient is prioritized, and you are actually displacing the benign patient."-*
> DIRECTOR SURGICAL DIVISION

Respondents noted that rationing strategies were used strategically to redirect patient flow. Hospitals focus their activities to more narrowly defined subpopulations or services. Consequently patients with low complexity needs (selection) were denied access, or hospitals used long waiting lists (delay) for low complexity services.

Rationing by selection was often interpreted as a strategy to improve patient care, rather than a method to cut costs. For most respondents it was difficult to discern efficiency measures from rationing. Besides, respondents found it hard to identify the direct consequences for their patient's health. Most respondents mentioned that competing hospitals had enough capacity to take over the patient flow. One potentially negative consequence for patients was increased travel time, and dissatisfaction due to their inability to go to the hospital of their first choice.

Discussion

This paper presents how Dutch hospitals have dealt with the introduction of six cost-increasing health technologies. The findings show that the opportunity costs of cost-increasing health technologies are not easily identifiable; limited transparency in the allocation of funds downstream within a hospital contributed to this. Furthermore, we found that the entry of new innovations and cost-containment are two parallel processes that are generally not causally linked. The way of financing is pivotal in displacement in the Netherlands, because there is a separate budget for expensive drugs. This budget pressure is reallocated horizontally across departments, whereas the budget pressure of remaining services is primarily reallocated vertically within departments or divisions. Hospitals have reacted to budget pressures primarily through a narrowing in the portfolio of their services, and a range of (other) efficiency measures. The board of directors is central in these processes, while insurers are involved only to a limited extent. Direct displacement of high-value care due to the introduction of new innovations was not observed. Rationing (primarily reducing accessibility) was observed mainly in response to cumulative cost pressures, production ceilings and capacity problems. Patients were prioritized on the basis of medical need, malign and acute patients were prioritized for benign and non-acute patients. It was hard to identify the direct consequences for patients' health.

Our analysis supports and builds on a relatively new field within health economics, a field that concerns identifying displacement effects as a response to the introduction of cost-increasing services, and estimating implicit threshold values to inform decision making concerning the basic benefit package. In line with Karlsberg Schaffer et al, we found that new technologies were generally accommodated by greater efficiency and increased spending, and that hospitals sought savings or efficiency measures in response to cumulative cost pressures rather than in response to single cost-increasing technologies.

One notable contribution of our research is that we, based on comparative analysis, identified two distinct pathways in which new technologies contribute to budget pressure. Financing is pivotal here. The first pathway includes funding for expensive drugs, which are explicitly appraised for inclusion in the basic benefit package. Once included, such drugs are generally generously reimbursed. This budget pressure is partly accommodated by insurers, and partly spread horizontally across several departments. The second pathway concerns funding for all other (non-pharmaceutical) technologies, which are rarely assessed by ZINL before entry, and the costs of which relate to the revenue ceiling. The budget pressure of such technologies is generally reallocated vertically within the department or division. Insurers only have limited abilities to control such spending, and hospitals have ample opportunity for cross-subsidization and to exchange funds. There is, however, generally a lack of clear-cut evidence about the value of the services. One risk in such implicit decision making processes is that policy be based on arguments that may not be in line with maximizing population health. Indeed, personal factors (e.g. the "powerful" medical doctor) and competition between providers were named as arguments for approving a business-case. In addition, the board of directors may have little insight into activities until costs escalate.

In England, Wales and Scotland, research into displacement has mainly focused on estimating the cost per QALY threshold [4,5]. If the objective is to maximize population health, the adoption of new technologies should depend on this threshold value, that equals the opportunity costs of marginal spending. Many questions remain about the assumptions underlying threshold estimations [15]. Such approaches vary in whether displacement is assumed to be optimal, for example that the least-cost effective program would represent the threshold, or the average ICER of all services would represent the threshold; and of the objective function of the reimbursement authority. Our approach is complementary to opportunity cost approaches. The opportunity cost approach answers the hypothetical question about benefits gained or lost due to alternative spending, while our research observes what happens in a system with a budgetary constraint when cost-increasing technologies enter this system. Our findings indicate that displacement typically does not take place at the level of individual technologies, such as assumed in QALY league table approaches. On the contrary, hospitals displace a range of low-value services, or services that may be provided elsewhere at lower costs, and decrease the volumes across the totality of their services.

Furthermore, our findings indicate that the opportunity costs vary across services. The budget pressure of expensive drugs is reallocated horizontally, and its opportunity costs may be equal to the marginal value of spending for the entire hospital. In contrast, the budget pressure of remaining services are predominantly reallocated vertically, and the opportunity costs thus depends on the efficiency of the particular department or division and the service they displace; i.e. the opportunity cost of LVAD (cardiothoracic and cardiology departments/ division) may differ from that of FEVAR (vascular surgery).

Implications for policy

Our findings indicate that rationing (primarily reducing accessibility) was observed mainly in response to cumulative cost pressures, production ceilings and capacity problems. Such problems are likely to worsen, given the newly established sector agreement with decreasing permitted budgetary growth (1.3% in 2019 to 0% in 2022, excluding wage and price adjustment). Active surveillance of waiting lists is warranted to prevent waiting list driven morbidity. Possibly, as cost constraints increase, more drastic approaches may be applied to accommodate new innovations, which may increase the opportunity costs of implementation. This implies that new technology assessment should not be viewed separately from the general budget constraints that hospitals face, and that in periods of increased cost-containment, new technology assessment might need to be stricter. Furthermore, we showed that – albeit many stakeholders are involved – the introduction of non-pharmaceuticals is relatively uncontrolled, and that this may have undesirable effects. Legislators might consider whether the 'open' description of the benefit package for non-pharmaceuticals could become more 'closed'. One option might be to extend managed entry agreement to non-pharmaceuticals. Besides, relevant stakeholders might join efforts to 'guide' the introduction of new innovations more prudently, for example through establishing minimum quality requirements. Finally, insurers might further develop their procurement policies to more effectively limit the entry of low-value innovations.

Strength and limitations

One major strength of our study is that we interviewed a wide range of stakeholders with diverse positions and responsibilities in the Dutch hospital sector. However, insurers were relatively underrepresented. For most respondents it was difficult to discern efficiency measures from rationing by dilution, and it proved hard to identify the direct consequences of rationing strategies. Other type of research may be needed to further study such effects. As with all qualitative research, our findings may not necessarily extend to other settings. We purposefully identified six contrasting case studies, other case studies may have led to other results. Our analyses primarily concerned academic or relatively large hospitals, as most innovations enter the sector in these hospitals. Besides, our findings are dependent on the Dutch local context, most notably the way Dutch hospitals are reimbursed. Social desirability or selective recall bias may have also influenced our findings. There may be no incentives for respondents to reveal displacement in interviews. However, our research methods were designed to cope with this problem, as we guaranteed that neither findings nor quotes would be attributed to individuals or organizations. Besides, we recruited a large number and diverse set of relevant stakeholders – also within hospitals – in order to verify and compare statements.

More research is needed to identify displacement mechanisms in healthcare domains other than the hospital sector. In addition, more research may be needed to further substantiate or adapt the currently held threshold values in the Netherlands. Our research identified two distinct pathways of displacement effects, which are intrinsically linked to financing, and our approach may be fruitful in other countries as well. Furthermore, our findings once more point to the plethora of low-value service provision and lack of knowledge of the value of many services. More research is warranted in disinvestment of low-value services, and of (early) health technology assessment to prevent the introduction of promising, but nevertheless low-value services.

Conclusion

The opportunity costs of cost-increasing health technologies are not easily identifiable. Hospitals typically displace a range of low-value services, or services that may be provided elsewhere at lower costs, and decrease the volumes across the totality of their services. The way of financing is pivotal in displacement in the Netherlands, as the budget pressure of expensive drugs is reallocated horizontally across departments, whereas the budget pressure of remaining services is primarily reallocated vertically within departments or divisions. Hospitals ration mainly in response to cumulative cost pressures, production ceilings and capacity problems, and active surveillance of waiting lists is warranted to prevent waiting list driven morbidity.

References

1 Zorginstituut Nederland, Pakketadvies Pembrolizumab. 2016.
2 Zorginstituut Nederland, Pakketadvies Pertuzumab. 2016.
3 Ministry of Health. Geslaagde prijsonderhandeling borstkankermiddel pertuzumab en longkankermiddel pembrolizumab. 2017 9 March 2018]; Available from: https://www.rijksoverheid.nl/actueel/nieuws/2017/06/09/geslaagde-prijsonderhandeling-borstkankermiddel-pertuzumab-en-longkankermiddel-pembrolizumab
4 Appleby, J., et al., Searching for cost effectiveness thresholds in the NHS. Health Policy, 2009. 91(3): p. 239-45.
5 Claxton, K., et al., Methods for the estimation of the National Institute for Health and Care Excellence cost-effectiveness threshold. Health Technol Assess, 2015. 19(14): p. 1-503, v-vi.
6 Karlsberg Schaffer, S., et al., Opportunity costs and local health service spending decisions: a qualitative study from Wales. BMC Health Serv Res, 2016. 16: p. 103.
7 Klein, R. and J. Maybin, THINKING ABOUT RATIONING. 2012, The King's Fund: London.
8 Kroneman, M., et al., The Netherlands: health system review. Vol. 18(2). 2016.
9 Mossialios, E., et al., 2015 International Profiles of Health Care Systems, C. Fund, Editor. 2016.
10 van der Wees, P.J., et al., The Relationship Between the Scope of Essential Health Benefits and Statutory Financing: An International Comparison Across Eight European Countries. Int J Health Policy Manag, 2015. 5(1): p. 13-22.
11 Raad voor de Volksgezondheid en Zorg, Zinnige en duurzame zorg. 2006: Zoetermeer.
12 Kroneman, M. and J.D. de Jong, The basic benefit package: composition and exceptions to the rules. A case study. Health Policy, 2015. 119(3): p. 245-51.
13 Baxter, P. and S. Jack, Qualitative case study methodology: study design and implementation for novice researchers, The Qualitative Report, Editor. 2008. p. 544-559.
14 Greuter, M.J.E., et al., Screening for Colorectal Cancer With Fecal Immunochemical Testing With and Without Postpolypectomy Surveillance Colonoscopy: A Cost-Effectiveness Analysis. Ann Intern Med, 2017. 167(8): p. 544-554.
15 Eckermann, S. and B. Pekarsky, Can the real opportunity cost stand up: displaced services, the straw man outside the room. Pharmacoeconomics, 2014. 32(4): p. 319-25.

Appendix 1 Interview scheme

Introduction

A consortium consisting of Radboudumc, Ecorys, Celsus, Maastricht University and Julius Center is conducting research into displacement effects in healthcare at the request of the National Health Care Institute. Displacement is described by the Healthcare Institute as follows: When assessing whether a treatment should be included in the insured package, it is assessed whether the health gain that can be achieved is in a reasonable proportion to the costs that have to be incurred (the cost-effectiveness). If this is not the case, the inclusion of the new treatment will be at the expense of the reimbursement of another treatment. This is based on the fact that the available money can only be used once. In order to map (potential) displacement at hospital level, we ask different care professionals and other stakeholders in the care for their opinion.

We would also like to discuss this with you during an interview of about 45-60 minutes. The goal is twofold:

- Find out what consequences the introduction of [case] has for your department and / or for the hospital and what choices were made as a result
- In addition, we are also curious about your vision of displacement in healthcare in general.

With your approval, the interview will be recorded using a voice recorder.

All data and information provided by you will be used confidentially and exclusively for the execution of this research. Do you have any questions or remarks so far?

Financing

First of all, we would like to gain insight into the way in which [case] is financed within your hospital.

- Could you please estimate the size of the patient group that is treated each year with [case] at your department / in the hospital?
- To what extent is [case] fully reimbursed by health insurers?
- Do you identify problems in the negotiation with health insurers about [case]?

Impact

- Has a horizon scan been carried out with regard to the introduction of [case] within your department / hospital?
- Has [case] led to identifiable problems at the ward / in the hospital?
- Has [case] led to certain choices that relate to regular care?
  - Do you recognize one or more rationing strategies [a table with the six strategies was given] as a result of [case]?
  - What do you think are the (possible) consequences of this rationing for patients?
  - To what extent has [case] influenced the possibility of (previously planned) investments at your department / in the hospital?
- What are your expectations regarding the future deployment of [case]?

Involved actors

Based on literature research and initial interviews, we have developed a model, in which we map the way in which displacement may take place in healthcare. I would like to discuss this model with you.

- [Presenting figure 1]
  - Who decides about the financing of [case] in your hospital?
  - Where is the decision to introduce [case] within your organization most felt and by whom specifically?
  - Do other hospitals in the region suffer from the choices that your hospital makes in relation to [case]?
  - Do you notice shifts in care at national level that occur as a result of the admission of [case]?
- Which external parties are possibly involved in displacement in healthcare and how?

Displacement in healthcare

- In your opinion has [case] led to the displacement of other care?
- Has it ever happened that, due to the budget pressure, you were unable to provide the care that you would like to? If so, what specifically could you no longer do or did not want?
- Can you think of an intervention that entails a high risk of displacement?

Closing

- Do you have any other information or documents that are relevant to this topic?
- Do you have any suggestions regarding other colleagues that we could discuss?
- In due course, may we approach you again for any additional questions if we are further in the study?
- Do you have any questions or comments?

Thank you for your time and effort.

Appendix 2 Results of the case-studies

Case study	Entry to the system	Reimbursement, contracts, negotiations	Difficulties or problems encountered	Decision making process and decisions, motivation, contra-mechanisms	Impact on regular care	Displacement?
Left ventricular assist device	Due to improvements in the service, indications for 'bridge' and 'destination' were not mutually exclusive anymore. Hospitals adopted destination therapy 1) Extending a end-stage heart failure program (starting in 2010) 2) Extending bridge to transplant program Consequently, they could treat patients that could otherwise not be treated due to a shortage of donor hearts. Departments or division wrote business-cases, or included growth of LVADs in their production plans for the coming year. Both were accepted by the board of directors. In 2015 the indication 'destination therapy' was approved for the benefit package.	Board of directors used to grant departments a budget for LVADs from innovation funds, on the condition that the departments would publish in academic journals about the LVADs and would ensure LVAD would be adopted in the basic benefit package. Later, LVADs were financed on the basis of fee for service, and carved out from the budget ceilings negotiations. Because LVADs are on list with 'non steerable' interventions, this led to no problems in negotiations. Prices are set nationally, no volumes are negotiated, but estimations are made in advance for planning (attract additional personal etc, or that insurers can anticipate for the extra budget).	Increased need for: - Specialized personnel. - Intensive care, cardiology beds. - Operation room capacity. Other organisational and medical problems.	Departments wrote business-cases for board of directors for more budget and personnel. National working group developed indication criteria and quality criteria. Motivation: LVADs are considered a priority topic by departments and hospitals. Contra-mechanisms: - Displace low complexity care from university hospital. - Efficiency measures, including reducing length of stay. - Negotiation to reduce the price of the LVAD.	**Delay**: other operative procedures are postponed. **Denial** of patients with low complexity needs.	No *financial* displacement within the hospital, mainly because of the financing arrangement with relatively low risk for providers. In addition, the growth in number of LVADs stayed behind expectations.

Case study	Entry to the system	Reimbursement, contracts, negotiations	Difficulties or problems encountered	Decision making process and decisions, motivation, contra-mechanisms	Impact on regular care	Displacement?
Fenestrated endovascular aneurysm repair	Specialists and departments started experimenting early 2000s, activities were scaled up after initial success. In 2013 FEVAR was approved for the benefit package. Some hospitals started with FEVAR after the arrival of a new professor with experience in FEVAR. Besides, the technology improves so that patients can be treated that could otherwise not be treated, and the technology is innovative/ technically challenging, which is appreciated by the medical staff. Competition between hospitals, hospitals are afraid to stay behind.	Some hospitals cross-subsidized FEVAR from other products. Prices and volumes are negotiated between hospital and insurer, but these are not binding, may be exchanged for any other care, as long as the total expenditures do not pass the negotiated budget ceiling. Thus, hospital and insurer primarily negotiate a total budget, which is secondarily substantiated by prices and volumes. Some hospitals did not have any contracts with insurers about FEVAR. Insurers rarely contract on basis of quality protocols, or review patient notes. Departments/division wrote business-cases generally in reaction to budget overruns, rather than proactively.	Budget overrun for departments, divisions, cardiovascular centre. FEVAR was *one of the* services contributing to the *cumulative* budget overrun. FEVAR was a minor contributor, other services contributed more significantly to the budget overrun. Increased need for facilities.	In some hospitals, extra budget was proactively negotiated within the department, division, hospital (overspend, from reserves) or from the insurer. In other hospitals, extra budget was negotiated (both with and without success) in response to the cumulative cost pressure. In one hospital, operation room time was reduced to limit the number of surgical procedures and spending. Motivation: - Aorta pathology is considered a priority topic by departments and hospitals. - Competition between providers. - Less invasive is the future for surgery. Contra-mechanisms: - Negotiate lower prices for stents. - Displace low complexity care from university hospital. - Efficiency measures, including reducing length of stay.	**Delay**: waiting lists for surgery. **Denial or deflection** of low complexity patients.	FEVAR was one of the services that contributed to cumulative cost pressures. In some hospitals, extra budget could be negotiated to accommodate this growth. In other hospitals, the growth of FEVAR was limited due to cost pressures from competing innovations. Lower prices for stents reduce the likelihood of displacement. Due to intransparent financing, no displacement effects could be traced.

Expensive oncolytics	Expensive oncolytics are reimbursed after EMA-registration, a positive evaluation of effectiveness by an established national oncologic commission ('Commissie BOM') and an assessment by the Health Care Institute. Within hospitals, drug committees discuss new drugs and negotiate extra budget from the board of directors.	Expensive oncolytics are reimbursed using add-on payments, most often on the basis of fee for service, or ex ante determined capped budgets. Hospitals negotiate with insurers about volumes and prices. In most hospitals, these negotiations are carved out from the negotiations guided by the budget ceilings. There are several requirements for carved out contracts. Insurers require that hospital adhere to guidelines, be transparent about costs and outcomes, and allow no margins on the drugs (which used to be the case, and was spent on other care).	Concerns about future financial sustainability, exorbitant high prices, and high profit margins of industry. Growth in spending for expensive oncolytics exceeds the 1% growth limit of sector agreements. Thus, growth allocated to expensive drugs needs to be retained from the other services.	Negotiate additional budget from insurers, before and during the year. Insurer and hospital first negotiate the total budget for the coming year (≈ budget-to +1%). Then this budget is cut to 1) all services that are carved out, and 2) other services, which are further cut in different segments of services. Board of directors may request departments to cut costs. Ongoing cycle of austerity/efficiency measures within the hospital, in line with the policy of the organisation. Motivation: - Insurers are afraid for loss of reputation. - Oncology is considered a priority topic in hospitals, malign is prioritized above benign. Contra-mechanisms: - Negotiate lower prices, through for example collective purchasing. - Precision medicine. - Efficiency measures. - Displace low complexity care from university hospital. - Introduction of new drugs delayed to next calendar year. - Cuts in (support) staff. - Cuts in number beds.	Rationing of expensive oncolytics was not reported, however **selection** (more strict adherence to indication criteria than before) was. Some treatments may be **delayed**, or patients were sent to other hospitals (**deflection**), however respondents were sure these had no negative impact on patient's health. Patients with an indication for expensive oncolytics were sent to and from other hospitals (**deflection**). Investments were delayed.	Expensive oncolytics lower the growth of remaining services. The budget pressure of the drugs was absorbed by board of directors and insurers, who redistributed this pressure to the rest of the departments. Because of this indirect route and intransparent financing, it is difficult to causally link austerity measures or budget cuts, to the budget pressure of the drugs.

Case study	Entry to the system	Reimbursement, contracts, negotiations	Difficulties or problems encountered	Decision making process and decisions, motivation, contra-mechanisms	Impact on regular care	Displacement?
Eylea and Lucentis	Lucentis has been reimbursed from 2007 and onwards. In 2012, ZINL advised to exclude Lucentis from coverage. In 2012 and 2014 respectively Lucentis and Eylea were included in the positive list for add-on payments. The drugs were initially indicated for patients with age-related macula edema. Eylea and Lucentis were also used for patients with diabetic macula edema and vascular occlusion. However, these indications were not included on the positive list for add-on payments until March 2015. When new indications appeared on the positive lists during the year, the board of directors tried to negotiate extra budget for the drugs with the health insurers.	Eylea and Lucentis are reimbursed using add-on payments, most often on the basis of carved out contracts. In some hospitals however, these services counted for the budget ceiling. In the past, eye injections were financed from DRGs (Avastin was mainly used for financial reasons) or from innovation funds. Insurers and providers negotiate a percentage of Eylea/Lucentis of total eye injections, typically between 10-20%. Insurers require transparency in the purchasing and use of the eye drugs.	Indication extension in the midst of the year led to budget overruns in hospitals where add-on payments for the drugs did count for the ceiled total budgets. Especially specialized eye centres experienced cost pressure due to expensive eye drugs. The number of patients and the percentage of patients requiring Eylea/Lucentis grows. For insurers it is difficult to determine the right percentage of expensive injections. Some insurers reimbursed more expensive eye injections than others. Increase in number of patients, increased workload.	Renegotiation with insurer. Internal redistribution of funds between departments by board of directors. Cross-subsidisation from other services with high profit margin. Board of directors control the volume and budget of departments. Motivation: - Board of directors decide about priority topics that are allocated a higher budget, compared to other departments/topics that are strictly kept to their budget. Contra-mechanisms: - Efficiency measures, including task rearrangements. - Displace low complexity care. - Improve adherence to indications, via committee for expensive drugs.	In exceptional situations, patients were **selected** on basis of insurance company. In addition, patients were selected on basis of medical urgency. **Deflection** of patients that need expensive drugs because of inadequate budget. **Delay** of non-acute care, or **denial** of referred patients. Under spend on innovations/ investments/ maintenance.	Specialized eye centres experienced most cost pressure, presumably because they had less abilities for cross-subsidiation and internal redistribution of funds, or had less market power to negotiate carved out FFS contracts for expensive drugs. Rationing by delay and selection were widely reported, however these were not causally linked to the eye drugs.

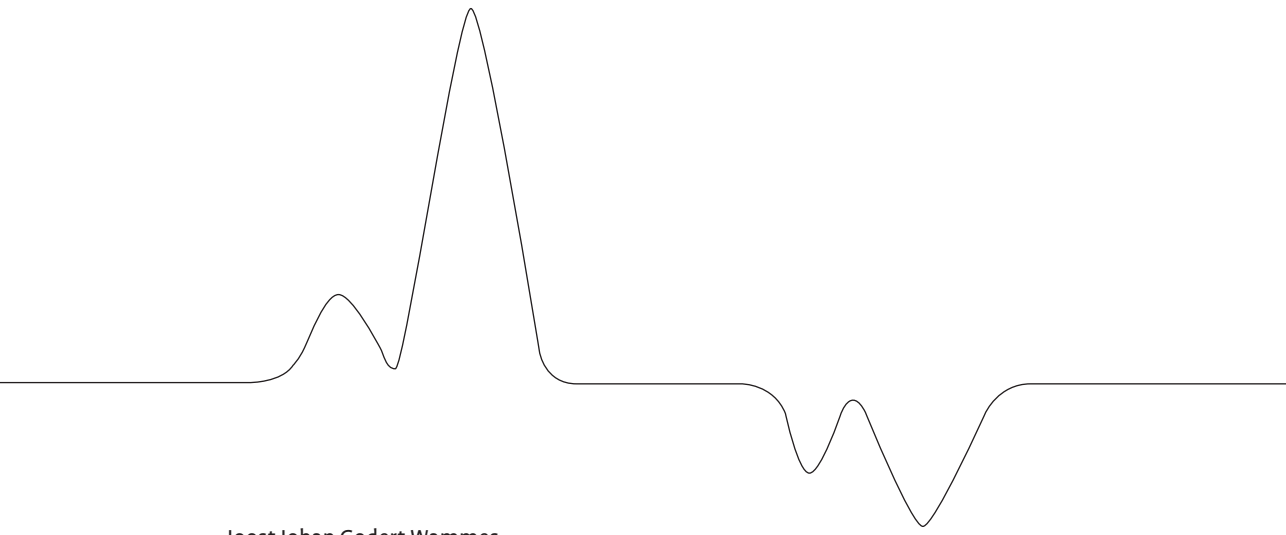
Population screening for colon cancer	Hospitals could choose to participate in the program. This was profitable due to 1) increased revenue for endoscopies 2) the extra patients because of resulting treatment of patients with positive test results. Hospitals are required to treat a minimum number of patients with colon cancer. Adoption of population screening in the benefit package was based on an advisory report of the Dutch Health Council.	Screening endoscopies were initially reimbursed per service, not counting for the budget ceiling. Later, hospitals and insurers explicitly negotiated about prices and volumes of screening endoscopies.	Undercapacity gastroenterology (GE) departments, shortage of GE-specialists. Higher demand for colonoscopies than anticipated.	Internal redistribution of funds between departments by board of directors. Departments and divisions are generally held responsible for their own budget and are given flexibility in how to spend the budget. Departments, divisions and board of directors determine priority topics for which budgets are loosened. Motivation: - Priority topic of departments and hospital. - Colon cancer and oncology in general were considered key topics in some hospitals. - Colon screening was considered low complexity care in some academic centres. - Colon cancer patients were generally prioritized above patients with inflammatory bowel disease. Contra-mechanisms: - Hospitals communicate their capacity for screening endoscopies digitally to the national program. As such, they were able to reduce the volume of services and demand locally. - Displace low complexity care. - Regional partnerships. - Substitution. - E-health.	**Deflection** of patients to other regions. **Delay**: waiting lists for regular endoscopies or GE-care in general. **Selection**: in urgent cases patients were given priority and were exempted from the waiting list. Various alternatives for selection based on urgency. **Denial** of request for second opinions. Academic hospitals deny low complexity patients.	Most of the reported problems or rationing were related to the pre-existing shortage of GE-specialists. The screening program put further pressure on the waiting lists, but the waiting lists were generally not linked to adverse health outcomes.

Case study	Entry to the system	Reimbursement, contracts, negotiations	Difficulties or problems encountered	Decision making process and decisions, motivation, contra-mechanisms	Impact on regular care	Displacement?
Robotic surgery	The first Da Vinci platform was bought in 2000. ZINL did assess Da Vinci, but not formally in- or excluded the service from the benefit package, because decisions about service coverage concern treatments for a given patient population, not the way the treatment is delivered. Within hospitals, departments (primarily urology) requested the board of directors to buy the platform.	The procurement of the platform is financed from investment- or innovation budgets. Insurers are not involved in and are not consulted for the purchase of the platform. Hospitals and insurers mainly negotiate budgets, and make no specific agreements about specific procedures. However, insurers may request a minimum number of procedures per year to guarantee high quality of care.	Lack of evidence for benefits of the platform. DRGs for the laparoscopic procedure are not sufficient to cover the costs. Higher reimbursement has been requested, but were rejected by the insurers because of the lack of evidence concerning the procedure. High price of the platform, because of the monopoly of the manufacturer. Intransparency of costs of procedures in the platform, compared to conventional procedures. In general: little insight in the costs of the organisation.	Medical specialists, urologists in particular, requested the board of directors to buy the platform, and wrote a business-case. Board of directors approved the request or not. Hospital tried to negotiate extra budget from insurers. The laparoscopic procedures are cross-subsidized from other funds or from services with profit margin in the hospital. Budget cuts for operation rooms, intensive care (primarily in budget for personnel). Reduction in procedures that were considered non priority topics of the hospital. Motivation: - Marketing/competition between providers, to attract patients, providers are afraid to lose prostate cancer patients.	**Delay**: waiting times for departments that are not considered key topics of the hospital. Malign was prioritized over benign. Opportunity costs for other investments/ innovations.	Lower use of the platform than anticipated has led to indication extension with questionable benefit for patients. Due to intransparency of costs the additional costs due to the platform are unknown. As a response to the budget pressure of the platform, hospital generally: reported the opportunity cost of other investments or innovation; prioritized some key topics above others; reduced investments

Lower use of the platform than anticipated. LIncreased costs of disposables.	- It was argued patients prefer robotic surgery over classical surgery. - Minimally invasive surgery was considered the future of surgery. - Oncology was considered a priority topic of the hospital. - Innovation/research was considered a core activity of the hospital. - Insurers aim to concentrate Da Vinci in a small number of high volume hospitals. - Larger hospitals generally have more abilities for cross-subsidisation and for equalisation, and stronger negotiation power. Contra-mechanisms: - Indication extension to recoup the purchase costs of the platform, or to attract patients in the region. - More strict adherence to indications. - Reduction in length of stay. - Increase in productivity. - Efficiency measures.	in personnel (operation room, intensive care).

CHAPTER 6

A systematic review of high-cost patients' characteristics and healthcare utilization



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Abstract

Objectives To investigate the characteristics and healthcare utilization of high-cost patients, and to compare high-cost patients across payers and countries.

Design Systematic review.

Data sources Pubmed and Embase databases were searched until October 30th, 2017.

Eligibility criteria and outcomes Our final search was built on three themes: 'high-cost', 'patients', and 'cost' and 'cost analysis'. We included articles that reported characteristics and utilization of the top-X% (e.g. top-5%, top-10%) patients of costs of a given population. Analyses were limited to studies that covered a broad range of services, across the continuum of care. Andersen's behavioral model was used to categorize characteristics and determinants into predisposing, enabling and need characteristics.

Results The studies pointed to a high prevalence of multiple (chronic) conditions to explain high-cost patients' utilization. Besides, we found a high prevalence of mental illness across all studies; and a prevalence higher than 30% in US Medicaid and total population studies. Furthermore, we found that high costs were associated with increasing age, but that still more than halve of high-cost patients were younger than 65. High costs were associated with higher incomes in the US, but with lower incomes elsewhere. Preventable spending was estimated at maximally ten percent of spending. The top-10%, top-5% and top-1% high-cost patients accounted for respectively 68%, 55%, and 24% of costs within a given year. Spending persistency varied between 24% and 48%. Finally, we found that no more than 30% of high-cost patients are in their last year of life.

Conclusions High-cost patients make up the sickest and most complex populations and their high utilization is primarily explained by high levels of chronic and mental illness. High-cost patients are diverse populations and vary across payer types and countries. Tailored interventions are needed to meet the needs of high-cost patients, and to avoid waste of scarce resources.

Key words health services administration and medicine; high-need high-cost; integrated delivery of health care; health care utilization, health care costs

Strengths and limitations of this study

- Based on an extensive literature search, this review included 55 studies of high-cost patients' characteristics and healthcare utilization.
- Andersen's behavioural model was used to categorize the characteristics of high-cost patients into predisposing, enabling and need characteristics.
- Grey literature was not included in our systematic review. However, we identified 55 studies and compared high-cost patients' characteristics and healthcare utilization across payers and countries.
- We did not assess the quality of the studies because of the methodological diversity of the studies.

Background

It is widely known that healthcare costs are concentrated among a small group of 'high-cost' patients [1]. Although they receive substantial care from multiple sources, critical health care needs are unmet, and many receive unnecessary and ineffective care [2-5]. This suggests that high-cost patients are a logical group to seek for quality improvement and cost reduction.

Especially in the US, many providers or insurance plans have pursued this logic and developed programs for "high-need, high-cost patients". So far, such programs, including for example care coordination and disease management, have had favorable results in quality of care and health outcomes, and mixed results in their ability to reduce hospital use and costs [6]. Research has shown that the effectiveness and efficiency of the programs increase when interventions are targeted to the patients that most likely benefit [2,7,8]. Little is known however, about variations in clinical characteristics and care-utilization patterns across payer-defined groups or countries [9]. Such insight in the health requirements of high-cost patients is prerequisite for designing effective policy or program responses.

We conducted this systematic review to synthesize the literature on high-cost patients' characteristics and healthcare utilization. Andersen's behavioral model (see method section) was used to organize the findings. Our analysis was aimed at identifying drivers of costs that matter across payer types and countries. We aimed to inform the development of new interventions and policy, as well as future research in high-cost patients.

Methods

Our methodology was based on established guidance for conducting systematic reviews [10,11]. Our main research questions was 'Who are the most expensive patients, what health care services do they use, what drives these high costs, and what drivers matter across payers and countries?'.

Study selection

A preliminary search in Pubmed was conducted to identify key articles and keywords. On the basis of these findings, we developed a search strategy covering the most important terms. We then reshaped the search strategy by consulting an information specialist of our university. The final search was built on three themes: 'high-cost', 'patients', and 'cost' and 'cost analysis'. The sensitivity of the search was verified with the key articles we found earlier. We searched Pubmed and Embase at October 30th, 2017. Full details of our search strategy are attached in appendix 1.

Inclusion and exclusion criteria

Articles were reviewed by Author A using title and abstract to identify potentially eligible studies. Author B verified a random sample of articles to guarantee specificity and sensitivity of the selection process. Only studies from high-income countries – as defined by the World Bank [12] – and studies published in 2000 and later were included. Studies not written in English and conference abstracts were excluded. In the second step, titles and

abstracts were reviewed by Author A to assess whether articles fit within our definition of high-cost patients: the article reported characteristics and utilization of the top-X% (e.g. top-5%, top-10%) patients of costs of a given population. Author B verified a random sample of articles at this selection step. In the third step, full-text articles were retrieved and independently screened by Author A and Author B for our inclusion criteria. At this step, we aimed for studies covering a broad range of services across the continuum of care at health system level, and excluded all studies with a narrow scope of costs (for example: hospital costs, pharmaceutical costs) and all studies with a narrow population base (primarily disease oriented studies, or studies in children). At each step of this selection process, (in-) consistencies were discussed until consensus was reached. On basis of the discussions, the criteria were refined and the prior selection process was repeated.

Data extraction

A data extraction form was developed by the research team to ensure the approach was consistent with the research question. Author A extracted all data. To guarantee specificity and sensitivity of data extraction, Author B and Author C both independently extracted the data of five random articles. A meeting was held to discuss (in-)consistencies in extraction results. On basis of this discussion, the data extraction form was refined and the prior data extraction was repeated. Per article the following key elements were extracted: author, year, country, definition of high-cost patients, in- and exclusion criteria of the study population, cost data used to determine total costs, characteristics of the high-cost patients such as diagnoses, age, gender, ethnicity, determinants for high costs including associated supply side factors (concerning the supply of health services), subpopulations, and health care use and costs (per subpopulation). We also made a narrative summary of the findings per article (provided in appendix 2). To identify the most important medical characteristics, only those diseases with a high prevalence (≥10%) among high-cost patient populations or medical characteristics overrepresented in high-cost populations were extracted. Medical characteristics (prevalent diseases) were categorized and presented at the level of ICD10-chapters.

Data synthesis

Andersen's behavioural model was used to categorize characteristics and determinants for high costs into predisposing, enabling and need characteristics. Andersen's model assumes that healthcare use is a function of 1) characteristics that predispose people to use or not to use services, although such characteristics are not directly responsible for use (e.g. age, gender, education, ethnicity, beliefs) 2) enabling characteristics that facilitate or impede use of services (income/wealth/insurance as ability to pay for services, organization of service provision, health policy) 3) needs or conditions that laypeople or health care providers recognize as requiring medical treatment. The model also distinguishes between individual and contextual (measured at aggregate level, such as measures of community characteristics) determinants of service use. Andersen hypothesized that the variables would have differential ability to explain care use, depending on the type of service. For example, dental care (and other discretionary services) would be explained by predisposing and enabling characteristics, whereas hospital care would primarily be explained by needs and demographic characteristics [13,14].

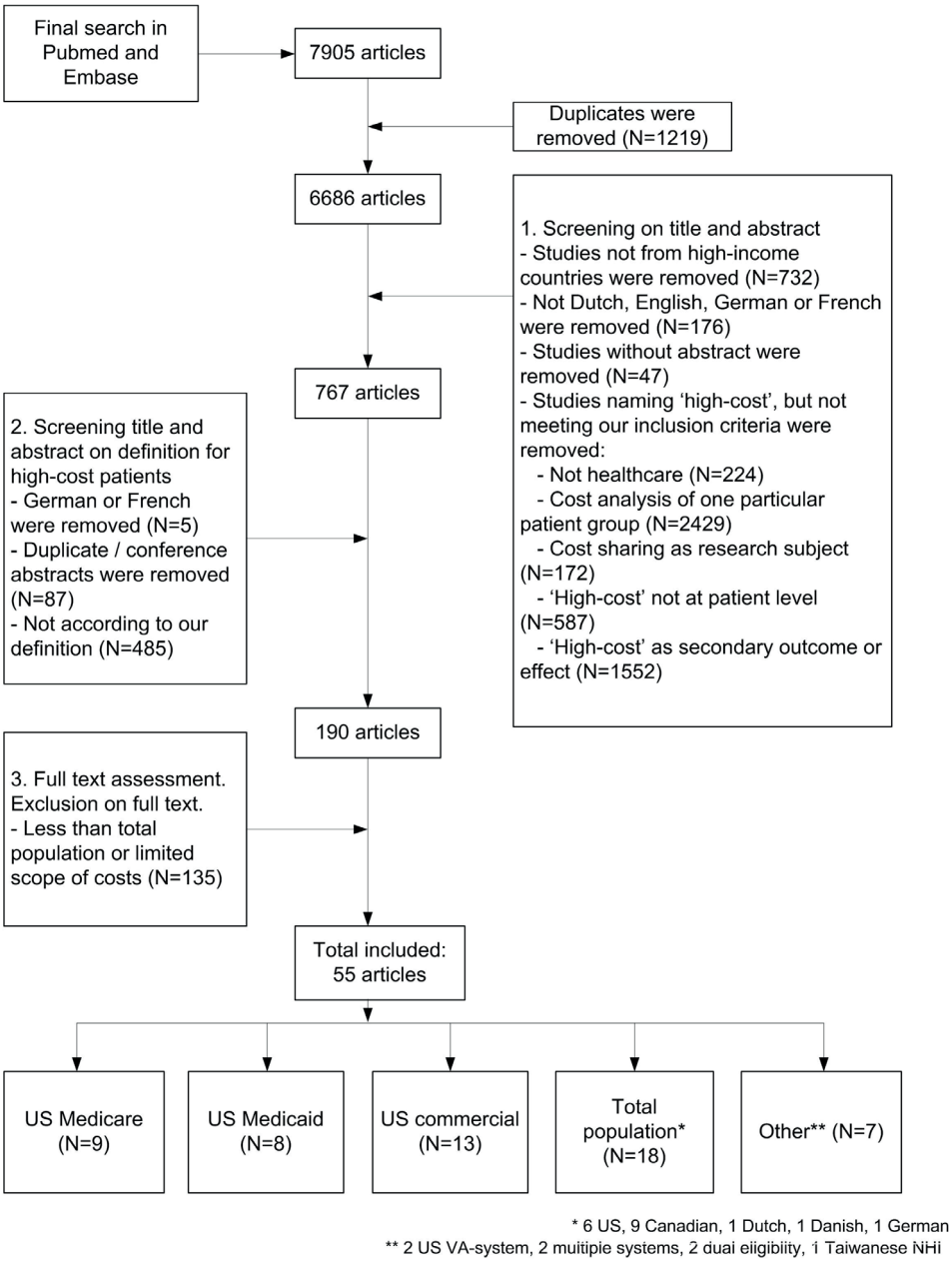


FIGURE 1 Selection process.

We presented all data according to five general categories, including study characteristics, predisposing characteristics, enabling characteristics, need characteristics, and expenditure categories and health care utilization. We presented summary tables of results, extracted central themes and topics from the studies, and summarized them narratively. All studies were analyzed according to payer and country to identify the most important drivers across settings.

Patient and Public Involvement

Patients and or public were not involved in the conduct of this study.

Results

General information

Our search strategy resulted in 7905 articles. After first broad eligibility assessment 767 articles remained. After screening of titles and abstracts, 190 articles remained for full-text screening, from which 55 were ultimately included (figure 1).

A description of the studies is given in table 1. The majority of the studies were conducted in the United States (N=42). The remaining studies were conducted in Canada (N=9), Germany (N=1), Denmark (N=1), the Netherlands (N=1), and Taiwan (N=1). All were retrospective cohort studies, and descriptive and logistic regression analysis were the main analytic approaches used. The study period ranged from six months to thirty years. The most frequent observation period was one year.

A range of definitions for high-cost patients were used, and some studies used more than one definition to distinguish between age groups, between high- and very high-cost patients, or to study persistently high-cost patients (>1 year high costs). In general, patients belonging to the top-1%, top-5%, top-10%, or top-20% of spending were considered high-cost patients.

The study population differed between the studies. We categorized eighteen studies as 'total population' studies, including studies in universal insurance schemes (of all ages; nine Canadian studies, one Dutch, one German, and one Danish study), studies that combined data of different payers, or survey studies. Respectively nine, seven and fourteen studies were among US Medicare, US Medicaid or US commercial populations. The remaining studies compared high-cost patients in multiple US payers, or were among US dual eligibles (eligible for both Medicare and Medicaid), US Veterans Affairs (VA)-beneficiaries, or among elderly in the Taiwanese insurance system. Some studies used additional criteria to determine the population. Age, healthcare use, or insurance were most frequently used as secondary condition to determine the population.

In fifty studies, total costs per patient were based on the insurance plan or public program. In the remaining studies, total costs were based on a survey or identified from a variety of sources.

TABLE 1 Description of the included studies.

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: in- and exclusion criteria	Cost data
Aldridge and Kelly[15], United States	Descriptive	2011	Top-5%	US population	Total spending was identified from a combination of data from MEPS, the Health and Retirement Study, peer reviewed literature, published reports, 2011 MEPS, 2011 National Health Expenditure Accounts.
Ash et al.[16], United States	Descriptive, logistic regression	1997-1998	Top-0.5% with highest predicted costs, top-0.5% prior cost.	Individuals eligible for at least one month in each of the two study years	MEDSTAT MarketScan Research Database, consisting of inpatient and outpatient care from individuals covered by employee-sponsored plans. Outpatient pharmacy costs were excluded.
Bayliss et al. [17], United States	Predictive modeling, cluster analysis	2014	Top-25%	Members with new Kaiser Permanente Colorado benefits and who completed the Brief Health Questionnaire	Per-member-per-month costs from Kaiser Permanente Colorado health system
Beaulieu et al. [18], United States	Descriptive, logistic regression	2011-2012	Top-10%	FFS Medicare population. Excluding patients <65 years, enrolled in Medicare advantage, and those not continuously enrolled in Parts A and B.	Standardized Medicare costs, excluding prescription drug charges.
Boscardin et al. [19], United States	Descriptive, logistic regression	2009	Top-10%	Employees enrolled in the Safeway health insurance program in 2009, with biometric and self-repored health status data (HRQ). Excluding: dependents covered through a family member.	Safeway's health plan
Buck et al. [20], United States	Descriptive	1995	Top-10%	Medicaid population in 10 states. Excluding: dually eligible, ≥65 years, enrolled in capitated plans, missing sex or birthdate.	Total Medicaid expenditures
Bynum et al. [21], United States	Descriptive, multinominal logistic regression	2010-2011	Top-10% in each state Persistently HC, died in 2011, or converted	Dually eligible adults with full Medicaid eligibility; in the 36 states that had usable and complete Medicaid data	Medicare and Medicaid

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: in- and exclusion criteria	Cost data
Chang et al.[22], United States	Descriptive, logistic regression	2007-2009	Consistent high-user: top-20% in four consecutive half year periods (=6.14% of the population) Point high-user: top-6.14% in 1 year	Enrollees from 4 health plans who were 1) continuously enrolled 2) incurred ≥$100 each year 3) 4 largest plan 4) aged between 18 and 62 in 2007. Excluding: those who died.	Commercial health plans
Charlson et al. [23], United States	Quantile regression	2007 (6 months)	Top-5%, top-10%	All enrollees of the MMC Plan who had an assigned primary care provider at Lincoln Medical and Mental Health Center.	Metroplus MMC costs, including inpatient, outpatient, ER, laboratory tests, and prescription drugs.
Charlson et al. [24], United States	Quantile regression	2009-2010	Top-5%, top-10%	Union of health and hospital workers in the Northeast, those who were consistently eligible for benefits over at least 22 months in 2009 and 2010 (self-insured trust fund), who also received DCG codes.	Inpatient, outpatient, emergency room, laboratory tests, behavioral health and prescription drugs.
Chechulin et al. [25], Canada	Logistic regression	2007/08-2010/11	Top-5%	All Ontario residents serviced by the Ontario healthcare system during the fiscal year 2009/10. Patients under five, or who died during this year were excluded	Total health system costs (including LTC), excluding outpatient oncology, outpatient dialysis, and outpatient clinic.
Cohen et al. [26], United States	Logistic regression	1996-2002	Top-10%,	Nationally representative sample of the Medical Expenditure Panel Survey	All direct payments to providers by individuals, private insurance, Medicare, Medicaid, and other payment sources for: inpatient and outpatient care; emergency room services, office-based medical provider services, home healthcare, prescription medicines, and other medical services and equipment.
Coughlin et al. [27], United States	Descriptive	2006-2007 (1 year)	Top-10%	Medicare beneficiaries and dual eligibles	Spending paid for by the public programs
Coughlin and Long [28], United States	Descriptive	2002-2004	Various. Top-1%, Top-5%, Top-10%, Top-25%, Top-50%	2002 national Medicaid population (living in institutions and community). Excluding: who received only SCHIP coverage or never full benefits. Top-0.1% of spenders.	Medicaid
Crawford et al. [29], United States	Neural network modeling	1999-2001	Top-15%	Members of a health plan, where American Healthways, inc. provided disease management services. Only members with 24 months continuous enrollment were included.	Health plan costs.

DeLia[30], United States	Descriptive, multinomial regression	2011-2014	Top-1%, top-2-10%, Persistently extreme: 4 years top-1% Persistently high: 4 years in top-10%	Medicaid/CHIP beneficiaries in New Jersey, newly covered individuals under the ACA (2014) were excluded, Medicaid/Medicare dual eligibles were excluded	Medicaid FFS claims and managed care encounters and CHIP
de Oliveira et al. [31], Canada	Descriptive	2012	Top-10%, top-5%, top-1%. Mental health HC patients: mental health >50% of total costs.	All adult patients (18 and older) who had at least 1 encounter with the Ontario health care system in 2012. Excluding: all individuals who did not have a valid Ontario Health Insurance Plan number.	Most publicly funded healthcare services.
Figueroa et al. [32], United States	Descriptive, chi-square	2012	Top-10%	Adults 18-64 year without FFS Medicare coverage or Medicare Advantage coverage.	Massachusetts All-Payer Claims database; nearly a universal account of all health care delivered in the state with the exception of Medicare FFS.
Figueroa et al. [33], United States	Descriptive	2012	Top-10%	All Medicare patients, excluding those with Medicare Advantage coverage, who were not continually enrolled in part A and B	Standardized Medicare costs.
Fitzpatrick et al. [34], Canada	Descriptive, logistic regression	2003/5 and five years follow up	Top-5%	Participants from two cycles of (CCHS) surveys, representative of the population ≥ 12 years and living in private dwellings. ≥ 18 years. Ex: baseline high-cost	Ontario health insurance plan
Fleishmann [35], United States	Logistic regression	1996-2003	Top-10%, top-5%	Nationally representative sample of the Medical Expenditure Panel Survey	All direct payments to providers by individuals, private insurance, Medicare, Medicaid, and other payment sources for: inpatient and outpatient care; emergency room services, office-based medical provider services, home healthcare, prescription medicines, and other medical services and equipment.
Ganguli et al. [36], United States	Descriptive, retrospective chart review, interview analysis	2005-2011	Five archetypal patients among the 50 costliest / 1500 highest cost patients	Patients selected by costs and a prospective risk score to participate in a Centers for Medicare and Medicaid care management project, >18 years and had sufficient cognitive capacity to participate in an interview, or if deceased had family members who were able to give sufficient information.	Total Medicare payments

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: in- and exclusion criteria	Cost data
Graven et al. [37], United States	Descriptive	2011-2013	Top-10%, Episodically high-cost, persistently high-cost	Adults ages 19 and over, enrolled in Oregon Medicaid, commercial or Medicare Advantage programs. Only those with continuous enrollment in 2011 and 2012 were included. Ex: dual eligibles, and individuals who had 'coordination of benefit'-claims or with negative total spending in any of the quarters.	Total Medicaid, commercial or Medicare Advantage payments (acute care expenditures), excluding spending on prescription drugs
Guilcher et al. [38], Canada	Descriptive	1 April 2010 – 31 March 2011	Top-5%	All persons eligible for provincial health insurance residing in the community, who had at least one interaction with the system in the last five years	All publicly funded healthcare in a universal public healthcare system
Guo et al. [39], United States	Descriptive, logistic regression	1999-2000	Top-10% of average monthly expenses	Medicaid, FFS recipients younger than 65. Excluding: nursing home recipients	Medicaid costs
Hartmann et al. [40], Germany	Logistic regression	2010-2011	Top-10%	Enrollees 18 years and older of AOK Lower Saxony, Germany's 10th-largest statutory health insurer	In- and outpatient care, sickness benefits, rehabilitation, home nursing, ambulatory drug supply, prescribed therapeutic appliances and remedies.
Hensel et al. [41], Canada	Descriptive, logistic regression	1 April 2011 – 31 March 2012	Top-1%, top-2-5%, top-6-50%, bottom-50%, and zero-cost referent group	All Ontario residents, with a valid Ontario health care, 18 years of age or older, and medical care costs greater than zero	Ontario health insurance plan, for all hospital and home care services, including physician care, costs related to outpatient physician services were not included
Hirth et al. [42], United States	Descriptive, logistic regression	2003-2008	High: top-10% Moderate: top-10%-30% Low: bottom-70% Usually low Low/moderate Sometimes high Often high Usually high	Under-65 population (Truven Health MarketScan database); enrollees and dependents of more than 100, mainly self-insured, medium and large employers Only people enrolled continuously are included. Attrition (a minority was enrolled each year) due to several reasons: death, retirement, children aging out of dependent status etc..	Data from all carve-outs (e.g., prescription drug, mental health), including claims for which the deductible is imposed. All spending was adjusted to 2008 dollars using the medical cost Consumer Price Index. Excluding: Out-of-plan spending (e.g., OTC drugs, travel costs).
Hunter et al. [43], United States	Descriptive, linear regression	Fiscal year 2010	Top-5%	Cohort from Veterans Affairs administrative records, who were eligible for and received care in study period. Ex: individuals with schizophrenia, bipolar depression, other psychosis, alcohol dependence and abuse, drug dependence and abuse, PTSD, and/or depression.	Inpatient, outpatient, pharmacy, and non-VA contract care.

Hwang et al. [44], United States	Descriptive, logistic regression	2008-2011	Top-10%	Employees from a large employer in Pennsylvenia and the employees' dependents. Only those continuously enrolled.	Amount paid by the insurer and the amount of cost sharing paid by individuals.
Izad Shenas [45], United States	Data mining techniques / predictive modeling	2006-2008	Top-5%, top-10%, top-20%	Nationally representative sample of the Medical Expenditure Panel Survey, household individuals ≥ 17 years (redundant records, or with zero personal-level weights were removed).	All direct payments to providers by individuals, private insurance, Medicare, Medicaid, and other payment sources for: inpatient and outpatient care; emergency room services, office-based medical provider services, home healthcare, prescription medicines, and other medical services and equipment.
Joynt et al. [46], United States	Descriptive	2011 and 2012	Top-10%	All Medicare patients, excluding those with Medicare Advantage coverage, who were not continually enrolled in part A and B, or who died during the study period	Standardized Medicare costs.
Joynt et al. [47], United States	Descriptive, linear regression	2009-2010	Top-10%	Medicare > 65 population. Ex: decedents, any Medicare advantage enrollment, not continuously enrolled.	In- and outpatient services.
Krause et al. [48], United States	Logistic regression	2009-2011	Top-5%, top-1%, >$100,000	Enrollees of Blue Cross Blue Shield of Texas, only members 18-63, with a zip code in Texas and continuous enrollment in 2009 were included.	Total claims expense, including expenditures for hospital care, outpatient facility services, and professional services.
Ku et al. [49], Taiwan	Descriptive, generalized estimating equations	2005-2009	Top-10%, top-11-25%	Survey respondents 65 years of age and older	National health insurance
Lauffenburger et al. [50], United States	Descriptive, group-based trajectory modeling	2009-2011	Top-5%	Patients ≥18 years, with continuous eligibility for the entire calendar year, with ≥1 calendar year before their entry year and with ≥1 medical and pharmacy claim in both the baseline and entry year.	Medical and prescription data of Aetna, a large US nationwide insurer
Lee et al., [51] United States	Descriptive, cluster analysis	2012	Top-10%	Medicare patients hospitalized exclusively at Cleveland Clinic Health System and received at least 90% of their primary care services at a CCHS facility	CCHS facility costs, post-acute care services were only included for those patients who were admitted to a CCHS post-acute care facility.
Leininger et al. [52], United States	Descriptive, logistic regression	2009-2010 (one year)	Top-10%	New enrollees for Medicaid who completed a self-reported health needs assessment	Medicaid costs.

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: in- and exclusion criteria	Cost data
Lieberman et al. [53], United States	Descriptive	1995-1999	Top-5%	Medicare FFS beneficiaries	Medicare spending
Meenan et al. [54], United States	Risk modeling.	1995-1996	Top-0.5%, top-1%	Enrollees of six HMOs, eligible for some period in 1995 and 1996, and who had an outpatient pharmacy benefit. Medicare Cost enrollees were excluded.	Total claims, including inpatient, outpatient, radiology, pharmacy, durable medical equipment, long-term care, laboratory.
Monheit [55], United States	Descriptive, logistic regression	1996-1997	Various. Top-1%, Top-2%, Top-5%, Top-10%, Top-20%, Top-30%, Top-50%.	Representation of non-institutionalized civilian US population (survey respondents)	Total payments (including OOP, uncovered services, third party payments).
Powers and Chaguturu[9], United States	Descriptive	2014	Top-1%	Patients of Partners HealthCare integrated delivery System	Medicare, Medicaid, commercial are compared
Pritchard et al. [56], United States	Descriptive	2011	Top-5%	Managed care population, of all ages, with at least 180 days continuous enrollment prior 1 January 2011, patients with gaps in enrollment greater than 30 days were excluded (so no uninsured or patients enrolled in traditional FFS Medicare or Medicaid programs)	Medical and pharmaceutical claims for more than 80 US health plans, the total amount reimbursed by the insurer plus the plan member's out-of-pocket share
Rais et al. [57], Canada	Descriptive	2009-2010 (1 year)	Top-5%	Cost consuming users of hospital and home care services at the provincial level.	Hospital and home care services. Excluding: Primary care and long term care use.
Reid et al. [58], Canada	Descriptive	1996-1997 (1 year)	Top-5%	≥18 years and older enrolled in the province's universal health care plan	Medical services costs in a universal health care plan (physician and hospital services)
Reschovsky et al. [59], United States	Descriptive, logistic regression	2006, or 12 months before death	Top-25%	Medicare FFS beneficiaries, ≥1 CTS survey, With USOC physician. Excluding: ESRD beneficiaries.	Standardized total costs of Medicare part A and B
Riley [60], United States	Descriptive	1975-2004	Top-1% Top-5%	Medicare, beneficiaries entitled to Part A and B	Medicare costs
Robst [61], United States	Descriptive, logistic regression	2005-2010	Top-1% in some years, or in six years	Medicaid beneficiaries with fee-for-service coverage for at least 6 months in all 6 years	Medicaid

Rosella et al. [62], Canada	Descriptive, multinomial logistic regression	2003-2008	Top-5% Top-1%, top-2%-5%, top-6-50%	Ontario residents. Participants of the CCH Survey. Excluding: Institutionalized. Full-time members of the Canadian forces. Persons living in remote areas/ aboriginal reserves. Ages 12-18.	Those covered by Ontario's Universal Health Insurance Plan (OHIP). Excluding: Some prescription drug costs, allied health services, dental care, eye care, assistive devices.
Snider et al. [63], United States	Logistic regression	2004-2009	Top-20%	Employees from large US employers, from the Thomson Reuters Marketscan Commercial Claims and Encounters database with both BMI and claims in any given year. Pregnant women and underweight employees were excluded.	All inpatient, outpatient, and prescription claims.
Tamang et al. [64], Denmark	Descriptive, prediction modeling	2004-2011	Top-10%	Entire population of Western Denmark, with a full year of active residency in year 1	Danish National Health Service
Wammes et al. [65], Netherlands	Descriptive	2013	Top-1%, top-2-5%, bottom-95%	Beneficiaries of one Dutch health insurer	Dutch curative health system, basic benefit package including voluntary complementary insurance benefits.
Wodchis et al. [66], Canada	Descriptive	April 1, 2009 – March 31, 2012	Top-1% Top-5% Top-10% Top-50%	People with a recorded age of less than 105 years who were alive on Apr. 1 in any of the three study years and who had a valid Ontario health care at any time between Apr. 1 2009 and March 31 2012.	Costs refer to health care expenditures that have been allocated to patient encounters for health care. All medically necessary care, both acute and long term, as covered by public health insurance. Excluding: Public health, community service agencies and many other programs, as well as for administrative (government) staff. Private home-care, privately insured medication costs.
Zhao et al. [67], United States	Descriptive, linear regression	1997-1999	Top-0.5%	Private insured, whose claims were covered in the Medstat MarketScan Research Database; a multi-source private sector healthcare database. All cases with a pharmacy benefit and at least one month of eligibility in each of the first two study years, or the last two study years.	Total medical costs, including inpatient plus ambulatory plus pharmacy costs, and deductibles, coinsurance and coordination-of-benefit payments.
Zulman et al. [68], United States	Descriptive, regression analyses	Fiscal year 2010	Top-5%	Veterans served by the VA System, who received inpatient or outpatient VA care.	Outpatient and inpatient, pharmacy, VA-sponsored contract care

Predisposing characteristics

Table 2 presents predisposing, enabling and need characteristics associated with high-cost patients. Age was related to high-cost patients in several ways. First, high-cost patients were generally older, and higher age was associated with high costs. This held for each payer type. Second, persistently high-cost patients were generally older than episodic high-cost patients, and higher ages were associated with persistently high costs. Third, the magnitude of cost concentration, and the threshold for high costs differed between age groups [66]. As younger groups are generally healthier, costs are concentrated among fewer individuals. Fourth, clinical diagnoses and utilization patterns varied across age groups [21,65,66], and some subgroups were related to particular ages, including mental health high-cost patients among younger ages [31]. Finally, although age was related to high costs, total population studies showed that approximately half of the high-cost populations were younger than 65 [38,65].

TABLE 2 Predisposing, enabling and need factors for high-cost patients.

Variables	Number of studies
Predisposing factors	
Age	32 [15, 17-20, 22, 25, 26, 28-30, 32, 34, 35, 37, 39-42, 47-50, 52, 55, 56, 58-60, 62, 63, 65]
Gender = male	9 [25, 30, 31, 39, 47, 51, 59, 61, 65]
Gender = female	16 [17, 19, 20, 26, 28, 30, 32, 37, 38, 42, 46, 55, 58, 62, 63, 65]
Ethnicity = black /African American	4 [18, 47, 59, 60]
Ethnicity = white	5 [20, 28, 34, 61, 62]
Ethnicity = less likely black or Hispanic	3 [28, 55, 61]
Ethnicity = less likely immigrant	1 [34]
Ethnicity = less likely whites	2 [46, 48]
Region	4 [28, 42, 45, 47]
Urban residence	6 [18, 38, 39, 46, 47, 49]
Rural residence	2 [25, 42]
Living institutionalized	3 [27, 30, 59]
Employment status: early retiree	1 [42]
Job satisfaction	1 [19]
Marital status: divorced/widow/separated/living alone	2 [26, 49]
Dependents less likely to incur high costs	1 [40]
Receive care in many census divisions	1 [59]
Harmful habits	3 [19, 52, 62]
Union membership	1 [42]
Education: less than a high-school degree (neigboorhod level)	1 [48]
Enabling factors	
Health insurance	
Medicare: more likely dual eligible	6 [18, 33, 46, 47, 59, 60]
Medicaid: specific eligibility status	4 [20, 28, 39, 61]
Commercial: increased insurance	2 [17, 42]
Total population: insurance status had no effect	1 [55]

Type of insurance	1 [40]
Income	
Positive relation with high costs	3 [26, 42, 55]
Negative relation	5 [25, 31, 34, 41, 58]
No relation	3 [49, 59, 62]
Organizational enabling factors	
Primary care physician supply	1 [47]
Specialist physician supply	1 [47]
Hospital bed supply	1 [47]
Medical specialist as usual source of care	1 [59]
Proportion of physicians who are medical specialists	2 [18, 59]
Inadequate time during office visits	1 [59]
Proportion of providers operating for profit	2 [18, 59]
Teaching hospitals	1 [18]
Low nurse-to-staffing ratios	1 [18]
Low supply of long term care beds	1 [18]
Regular medical doctor or hospital	1 [52]
Regular medical doctor (negative relation)	1 [62]

Need factors	
A00–B99 Certain infectious and parasitic diseases	9 [22, 26, 30, 34, 42, 61, 63, 65, 66]
C00–D48 Neoplasms	21 [18, 25, 34, 37, 42-44, 46-51, 56-58, 60, 63, 65, 66, 68]
D50–D89 Diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism	4 [21, 30, 56, 58]
E00–E90 Endocrine, nutritional and metabolic diseases	32 [16-19, 21, 22, 25, 27-30, 32, 34, 37, 39, 40, 43, 44, 46, 47, 49-53, 58, 60, 63-65, 67, 68]
F00–F99 Mental and behavioral disorders	34 [9, 18-23, 25, 27, 28, 30, 31, 33, 34, 37, 39-43, 46, 47, 50-53, 57, 58, 60-62, 65, 66, 68]
G00–G99 Diseases of the nervous system	10 [22, 30, 44, 46, 57, 58, 61, 63-65]
H00–H59 Diseases of the eye and adnexa	5 [34, 39, 57, 58, 65]
I00–I99 Diseases of the circulatory system	36 [9, 16-19, 21, 22, 25, 27, 29-31, 33, 34, 37, 40, 42-44, 46-53, 56-58, 60, 64-68]
J00–J99 Diseases of the respiratory system	30 [9, 16-18, 21, 22, 25, 26, 28, 30, 32, 34, 37, 39, 40, 43, 44, 46, 47, 49-52, 57, 58, 60, 64-67]
K00–K93 Diseases of the digestive system	9 [30, 31, 34, 42, 43, 57, 58, 61, 65]
L00–L99 Diseases of the skin and subcutaneous tissue	5 [30, 34, 39, 58, 65]
M00–M99 Diseases of the musculoskeletal system and connective tissue	15 [9, 18, 19, 22, 30, 34, 42, 43, 46, 50, 51, 56, 58, 65, 68]
N00–N99 Diseases of the genitourinary system	22 [9, 18, 21, 25, 30, 32, 34, 37, 40, 42-44, 46, 47, 49, 51, 56-58, 60, 64, 65]
O00–O99 Pregnancy, childbirth and the puerperium	5 [23, 33, 39, 58, 66]
Q00–Q99 Congenital malformations, deformations and chromosomal abnormalities	1 [64]
R00–R99 Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	6 [19, 34, 39, 51, 58, 65]

S00–T98 Injury, poisoning and certain other consequences of external causes	9 [34, 39, 42, 46, 48, 51, 57, 65, 66]
Z00–Z99 Factors influencing health status and contact with health services	3 [34, 57, 65]
Chronic illness	22 [15, 17, 18, 30, 32, 33, 35, 37, 39, 40, 43, 46, 49, 50, 53, 58, 60, 62, 64-66, 68]
Multimorbidity / burden of comorbid illness	31 [9, 15, 16, 19, 23, 24, 29, 30, 33, 35, 37-40, 42, 43, 45-47, 50, 56, 58-63, 65, 67, 68]
Decedents / survival	14 [15, 21, 30, 38, 39, 46, 53, 55, 58-60, 64-66]
Activities daily living	7 [17, 26, 27, 35, 45, 49, 55]
Health status	9 [17, 26, 35, 44, 45, 49, 53, 55, 62]

Studies showed inconsistent results for gender. Respectively 9 and 16 studies noted males and females were overrepresented in high-cost patients. Besides, gender was associated with different segments of the high-cost population, including males in top-1% or persistently extreme-cost patients, and females in top-2-5% or persistently high-cost patients [30,65], or males in mental health high-cost patients [31].

Eleven studies reported the association between ethnicity and high costs. In two Canadian total population studies and three US Medicaid studies whites were overrepresented among high-cost populations, whereas in four US Medicare studies Blacks were overrepresented.

Socioeconomic status is regarded as both a predisposing characteristic and an enabling characteristic in Andersen's model, and we found evidence for both relationships. One Canadian study found that high costs were most strongly associated with food insecurity, lower personal income, non-homeownership and living in highly deprived or low ethnic concentration neighborhoods [34]. Other studies found that social deprivation seemed to increase risk for high costs more than material deprivation [25].

Ganguli et al studied health beliefs among high-cost US Medicare patients: socioeconomic status, social network, patient activation, and relationships with and trust in the clinician and the health system all increased or decreased costs, depending on the context. Trust was particularly important, and modified the interaction between patient activation and costs: when patients trusted their physicians, patient activation was associated with lower costs. When trust was lacking, patient activation was associated with higher costs [36].

Health behaviors, including underweight, obesity, physical inactivity and former smoking were significantly related to high costs [62,63].

Enabling characteristics

The studies' abilities to assess the effect of insurance were limited because most study populations were determined by insurance. Nevertheless, the studies indicated that increased insurance may have indicated specific or additional care needs. For example, six US Medicare studies reported that high-cost patients were more likely dually eligible and four US Medicaid studies reported that certain eligibility statuses were associated with high costs. In addition, increased insurance was associated with high costs because it lowers costs. Two US commercial studies mentioned that high-cost patients were more likely to have a health maintenance organization plan, a preferred provider organization plan, or

comprehensive insurance compared to high-deductible health plans; and insured status was associated with less consideration of costs in decision making [36].

Twelve studies addressed the relationship between income and high costs. In three US studies higher incomes were associated with high costs, whereas five Canadian studies found that lower incomes were associated with (mental health) high costs. However, one US, one Taiwanese, and one Canadian study reported that income was not significantly related to high costs. Finally, among high-cost US Medicare patients, personal resources and education were associated with increased use of resources (higher SES was linked to higher priced care), but also with lower resources use [36].

Organizational enabling factors

The number of primary care physicians, specialists and hospital beds were associated with higher per capita preventable costs among high-cost US Medicare patients [47]. Reschovsky et al found several weak or insignificant relationships between organizational factors and high costs within the high-cost population, but found that high-cost US Medicare patients more likely had a medical specialist as usual source of care than a primary care physician or surgeon [59]. Finally, high-cost US Medicare patients were only modestly concentrated in hospitals and markets (they were widely distributed through the system). High concentration hospitals (with relatively many high-cost patients) had a 15% higher median cost per claim, were more likely for-profit and teaching hospitals, had lower nurse-to-patient ratios, were more likely to care for the poor, and had higher 30-day readmission rates and lower 30-day mortality rates. High concentration hospital referral regions had higher annual median costs per beneficiary, a larger supply of specialists but equal supply of total physicians, a lower supply of long term care beds, higher hospital care intensity and higher end-of-life spending [18].

Need characteristics

Medical characteristics of high-cost patients are presented in table 2. We categorized medical characteristics to ICD10-chapters. Circulatory diseases, mental and behavioral disorders, endocrine, nutritional and metabolic, diseases of the respiratory system, diseases of the genitourinary system, neoplasms and diseases of the musculoskeletal system and connective tissue were most frequently reported among high-cost patients. The prevalence of chronic disease(s) and multimorbidity were also dominant among high-cost patients. For example, Bynum et al showed that over 26.4% of high-cost US dual eligibles suffered from five or more chronic conditions [21].

Two studies presented medical characteristics across US payers. Both studies showed that high-cost commercial patients had the lowest numbers of comorbidities and that high-cost Medicaid patients had the highest prevalence of mental illness [9,37]. We further compared the prevalence of diabetes, congestive heart failure, lung disease, and mental disorders across the studies. The prevalence of diabetes, congestive heart failure and lung disease was relatively low (≈5%-25%) in US commercial and total population studies. In US Medicaid, the prevalence of congestive heart failure and lung disease were relatively high (≈15%-40%; one study reported a prevalence of diabetes and lung disease > 60% [32]), and the prevalence of mental illness was particularly high (≈30%-75%). In US Medicare, the

prevalence of diabetes, congestive heart failure and lung disease were highest (≈20%-55%) and the prevalence of mental illness more modest (≈10%-25%). In total populations, approximately 30-40% of high-cost patients were treated for mental illness. Besides, the prevalence of each of the chronic diseases in the Dutch study was comparable with the prevalence in other total population studies. Finally, persistent high-cost patients had a higher number of comorbidities and a higher prevalence of each of the diseases compared to episodic high-cost patients.

High-cost patients were more likely to die, and those in the process of dying were more likely to incur high costs. The mortality differed between payers, much less between countries. The mortality among Danish and Dutch high-cost patients was comparable with the mortality in other total population studies. In US Medicare studies the mortality ranged from 14.2% to 27.4%, compared to 11.7% in one US Medicaid study and 5% to 13% in total populations. In addition, top-1% patients were more likely to die compared to top-5% patients [55,65] and persistent high-cost patients were more likely to die than episodic high-cost patients [64]. Finally, among US dual eligibles, mortality varied much across age and residence groups; nearly half of dual eligibles aged 65 and older died [21].

Expenditure patterns and healthcare utilization

In each study, costs were heavily concentrated. The top-10% patients roughly accounted for about 68% of costs (range: 55%-77%), the top-5% patients accounted for about 55% of costs (range: 29%-65%) and top-1% patients for approximately 24% (range: 14%-33%) within a given year. Costs were generally less concentrated in US Medicare, and more concentrated in total populations.

A wide range of parameters were used to describe high-cost patients' healthcare utilization (table 3). Inpatient acute hospital care was most often reported as a primary expenditure category for high-cost patients. In line with this, seventeen studies reported hospitalizations, admissions or inpatient days as important cost drivers. Lieberman found that total spending per beneficiary correlated strongly with the use of inpatient services [53], likewise several studies found that increasing levels of use (i.e. top-1% compared to top-5%) were associated with increasing proportions of spending on (inpatient) hospital care [36,49,56,62,65,66]. Guo et al reported that high-cost users consumed more units of each of the service category analyzed, with the exception of laboratory tests [39]; these findings were confirmed elsewhere [44,56]. In addition, it was found that 91% of high-cost patients received care in multiple care types [57]. Mental care services were listed as expenditure category only in studies of total populations, US Medicaid, and US VA. Finally, one study determined the frequency use of expensive services among high-cost patients: expensive treatments (expensive drugs, intensive care unit treatment, dialysis, transplant care, and DRGs >€30,000) contributed to high cost in approximately one third of top-1% patients, and in less than ten percent of top-2-5% patients [65].

TABLE 3 Expenditure patterns and utilization of high-cost patients.

	NUMBER OF STUDIES
Spending category	
(Inpatient) hospital care	31 [18, 19, 21, 25, 27-29, 31-33, 36-39, 43, 44, 46, 49, 51-53, 56, 57, 59, 60, 62-66, 68]
Subacute care / postacute care services rehabilitation	11 [9, 25, 27, 28, 32, 33, 46, 56, 57, 59, 66]
Hospitalizations/ admission / patient days/ length of stay	17 [19, 29, 31, 33, 36, 38, 39, 43, 45, 47, 50-52, 56, 58, 65, 68]
Emergency department	12 [19, 37-39, 43, 44, 47, 50, 51, 56, 57, 68]
Outpatient (physician) visits	13 [26, 33, 38, 39, 43, 44, 49, 50, 56, 59-61, 68]
Long term care	11 [21, 25, 27, 28, 32, 33, 40, 43, 51, 61, 66]
Mental health	10 [20, 25, 28, 31, 39, 43, 57, 61, 65, 68]
Physician services	13 [29, 31, 39, 43-45, 56, 58-61, 66, 68]
Intensive care unit	2 [51, 65]
Prescription drugs	16 [22, 26, 28, 29, 32, 36, 38, 39, 44, 46, 50-52, 56, 65, 68]
Persistency	
Subsequent use	13 [21, 22, 28, 30, 34, 36, 37, 42, 53, 55, 60, 61, 64]
Prior use	5 [16, 19, 26, 34, 64]
Persistent users	21 [15, 16, 19, 21, 22, 25, 26, 28, 30, 34, 36, 37, 42, 44, 47, 53, 55, 60, 61, 64, 66]
Prediction of high-cost patients[1]	16 [16, 17, 19, 23-26, 29, 35, 40, 50, 52, 54, 61, 63, 67]

[1] An in-depth discussion of prediction models for high costs is beyond the scope of the article (though individual predictors are used throughout the paper). Generally, diagnosis based models outperform prior cost models, and combinations accurately predict high-cost patients. Besides, comorbidity indices also accurately predict high-cost patients, and self-reported health data meaningfully improved existing models.

Four studies quantified the amount of 'preventable' spending (based on preventable emergency department visits and preventable (re-)admissions) among high-cost patients. As shown above, various supply side characteristics were associated with higher preventable costs among high-cost US Medicare patients, and approximately 10% of total costs were preventable [47]. Another study found that 4.8% of US Medicare spending was preventable, and that high-cost patients accounted for 73.8% of preventable spending. Moreover, 43.8% of preventable spending was accounted for by frail elderly, and preventable spending was particularly high for heart failure, pneumonia, COPD/asthma and urinary tract infections [33]. Figueroa et al found that preventable spending differed by insurance type among US non-elderly: respectively 3,5%, 2.8% and 1.4% of spending were preventable among US Medicaid, US Medicaid managed care and privately insured high-cost patients [32]. Similarly, Graven et al found that proportions of preventable spending differed between payers, and that persistent high-cost patients had higher proportions of preventable spending [37].

Twenty-one studies reported on the persistency of high costs. We found three approaches for studying persistency. First, studies reported prior healthcare use and/or reported posterior healthcare use for patients with high costs in a given index year. In other studies, persistent high-cost patients were compared to episodic high-cost patients. Spending persistency varied between 24% and 48% for top-5% patients, and between 28% and 45% for top-10% patients. Spending persistence was relatively high in US Medicaid, and relatively low in US Medicare. Increasing persistence was associated with increasing expenditures on all service types [44].

Discussion

We reviewed 55 studies on high-cost patients' characteristics and healthcare utilization, and made comparisons across payers and countries. The studies consistently point to a high prevalence of multiple (chronic) conditions to explain high-cost patients' utilization. Besides, we found a high prevalence of mental illness across all the studies, most notably in US Medicaid and total population studies. We found that various health system characteristics may contribute to high costs. Preventable spending was estimated at maximally ten percent of spending. Furthermore, we found that high costs are associated with increasing age and that clinical diagnoses and utilization patterns varied across age groups. However, still more than half of high-cost patients are younger than 65 years. High costs were associated with higher incomes in the US, but with lower incomes elsewhere. Finally, we confirmed that high-cost patients are more likely to die, and decedents are more likely to incur high-costs. However, no more than 30% of high-cost patients were in their last year of life.

Strengths and weaknesses

This is the first systematic review of scientific literature on high-cost patients' characteristics and healthcare utilization. Future studies might consider inclusion of grey literature. We included studies of various payer types and countries, allowing comparisons across settings. However, most studies were conducted in the United States and Canada, which limits the generalisability of the findings. Although our comparison across countries did not reveal large differences in mortality or prevalence of common chronic diseases, these analyses were based on a limited number of variables, studies and countries. It is likely that the specific characteristics and utilization of high-cost patients vary across localizations due to a wide range of epidemiological and health system factors. One limitation is that we, because of methodological diversity, did not assess the quality of the included studies, and some studies by design did not control for confounding. To our knowledge, no agreed upon framework exists for risk of bias assessment of the kind of studies included in our review. One limitation in current frameworks for observation/cross-sectional studies is that these are primarily designed for studies that aim to assess intervention effects in comparative studies. The internal validity of the findings in our included studies is mainly contingent upon its ability to control for relevant confounders. However, no consensus exists about what factors should reasonably be controlled for. The external validity of the findings of each of the studies depend upon the breadth of the population studied, and the scope of the costs

considered for establishing total costs. Our study selection process was aimed at identifying studies with a broad population studied, and a wide range of costs considered. Finally, the studies used various approaches for defining the needs and measuring multimorbidity among their populations, which limits the comparability across studies.

Reflections on our findings

Current research in high-cost patients has focused on care redesign of the treatment of patients with multiple chronic morbidities [7,69]. One contribution of our review is our identification of notable differences in characteristics and utilization across payers and countries. This (clinical) diversity of high-cost patients may even be larger at a local level. Segmentation analysis has been suggested as a method to identify homogenous and meaningful segments of patients with similar characteristics, needs and behavior, that allows for tailored policy [70]. Such segmentation analysis may powerfully inform population health management initiatives. Given the multiple needs and cross-sectoral utilization of high-cost patients, we suggest such analyses should capture both characteristics and utilization as broadly as possible, to fully apprehend high-cost patients care needs and utilization. In the context of high-cost patients, multimorbidity complicates segmentation, and the usefulness of segmentation may depend on the way multimorbidity is dealt with. To illustrate a potent example, Hayes et al defined high-need, high-cost patients as "people having three or more chronic conditions and a functional limitation that makes it hard for them to perform basic daily tasks" [71].

Our findings also reveal several supply side factors that contribute to high costs. However, no firm conclusions can be drawn about the strength of these effects. The apparent limited impact of organizational factors on spending is in line with Andersen's model predictions, where multimorbidity and health status are prime determinants of healthcare costs [72]. However, such findings are surprising given the abundance of evidence for supplier induced demand and medical practice variation [73]. High-cost populations may be too diverse for studying the impact of organizational factors; for such studies more homogenous populations may be prerequisite.

Four of our included studies estimated the amount of 'preventable' spending among high-cost patients. Preventable spending was estimated at maximally ten percent of spending, which is relatively low compared to the amounts of savings that have been reported elsewhere [8]. Preventable spending was mainly defined as preventable emergency department visits or preventable (re-)admissions, as such echoing the two primary targets of most high-need high-cost programs, including care coordination and disease management. The algorithms used were said to be relatively narrow and could have included other diagnostic categories [37]. Besides, future studies might consider more broad measures of preventable or wasteful spending, and develop algorithms to identify duplicate services, contra-indicated care, unnecessary laboratory testing, unnecessary prolonged hospitalizations, or any other kinds of lower value services.

It was striking that three US studies reported that higher incomes were associated with high costs, whereas other studies found that lower incomes were associated with high costs. These findings may point to disparities in health, the price that some Americans pay for their care, and the reduced accessibility to care of low income patients. This may particularly hold

for the uninsured. Besides, these findings suggest tailored interventions for lower income patients may be worthwhile.

Policy and research implications

Based on our findings, we deduced four major segments of high-cost patients for which separate policy may be warranted, including patients in their last year of life, patients experiencing a significant health event who return to stable health (episodically high-cost patients), patients with mental illness, and patients with persistently high costs characterized by chronic conditions, functional limitations and elder age.

Many interventions have been taken to increase value of end-of-life care. Advance care planning has shown to increase the quality of end-of-life care and decrease costs [74-76]. In addition, health systems might consider strengthening their palliative care systems [77]. Increasing value for episodically high-cost patients requires appropriate pricing of procedures and drugs, for example through selective contracting of providers, reference pricing or competitive bidding [78]. In addition, bundled payments for procedures and associated care may improve care coordination and reduce the use of duplicative or unnecessary services [79]. Multidisciplinary needs assessment and shared decision making may reduce unwarranted variation in expensive procedures. Mental health high-cost patients are known for their medical comorbidities, which suggests these patients might benefit from multidisciplinary cross-sectoral healthcare delivery, for example through collaborative care [80,81]. Finally, persistent high-cost patients might benefit from a variety of models, including disease management, care coordination, or ambulatory intensive care units, depending on the needs of the population and local circumstances [8,82-84]. Especially population health management approaches may be beneficial for these populations. Sherry et al. recently examined five community-oriented programs that successfully improved care for high-need, high-cost patients. The five programs shared common attributes, including a 'whole person' orientation, shared leadership, flexible financing and shared cross-system governance structures [85].

One study addressed health beliefs and patient networks among high-cost patients [36]. More of such research is needed as health beliefs may be more amenable to change than other drivers of high costs. One study analyzed the use of expensive treatments by high-cost patients [65]. Better insight in such healthcare utilization patterns is needed to inform interventions and policy aimed at high-cost populations. There is a need for segmentation variables and logic that is informative at either micro-, meso- and macrolevel. More research is needed to identify determinants of preventable and wasteful spending.

In conclusion, high-cost patients make up the sickest and most complex populations and their high utilization is primarily explained by high levels of chronic and mental illness. High-cost patients are diverse populations and vary across payer types and countries. Tailored interventions are needed to meet the needs of high-cost patients, and to avoid waste of scarce resources.

References

1 Zook, C.J. and F.D. Moore, *High-cost users of medical care*. N Engl J Med, 1980. 302(18): p. 996-1002.
2 Blumenthal, D., et al., *Caring for High-Need, High-Cost Patients – An Urgent Priority*. N Engl J Med, 2016. 375(10): p. 909-11.
3 Bodenheimer, T. and A. Fernandez, *High and rising health care costs. Part 4: can costs be controlled while preserving quality?* Ann Intern Med, 2005. 143(1): p. 26-31.
4 Colla, C.H., et al., *Association Between Medicare Accountable Care Organization Implementation and Spending Among Clinically Vulnerable Beneficiaries*. JAMA Intern Med, 2016. 176(8): p. 1167-75.
5 Wennberg, J.E., et al., *Inpatient care intensity and patients' ratings of their hospital experiences*. Health Aff (Millwood), 2009. 28(1): p. 103-12.
6 Blumenthal, D., et al., *Tailoring Complex-Care Management, Coordination, and Integration for High-Need, High-Cost Patients, 2016., in Vital Directions for Health and Health Care Series.* , Discussion paper. National Academy of Medicine. Washington DC., Editor. 2016.
7 Anderson, G.F., et al., *Attributes common to programs that successfully treat high-need, high-cost individuals*. Am J Manag Care, 2015. 21(11): p. e597-600.
8 Brown, R.S., et al., *Six features of Medicare coordinated care demonstration programs that cut hospital admissions of high-risk patients*. Health Aff (Millwood), 2012. 31(6): p. 1156-66.
9 Powers, B.W. and S.K. Chaguturu, *ACOs and High-Cost Patients*. N Engl J Med, 2016. 374(3): p. 203-5.
10 Liberati, A., et al., *The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: explanation and elaboration*. BMJ, 2009. 339: p. b2700.
11 Moher, D., et al., *Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement*. BMJ, 2009. 339: p. b2535.
12 World bank. [cited 2017; Available from: http://en.wikipedia.org/wiki/World_Bank_high-income_economy
13 Andersen, R.M., *Revisiting the behavioral model and access to medical care: does it matter?* J Health Soc Behav, 1995. 36(1): p. 1-10.
14 Kominski, G.F., *Changing the U.S. Health Care System: Key Issues in Health Services Policy and Management*. 2013: Wiley.
15 Aldridge, M.D. and A.S. Kelley, T*he Myth Regarding the High Cost of End-of-Life Care*. American journal of public health, 2015. 105(12): p. 2411-2415.
16 Ash, A.S., et al., F*inding future high-cost cases: comparing prior cost versus diagnosis-based methods*. Health Serv Res, 2001. 36(6 Pt 2): p. 194-206.
17 Bayliss, E.A., et al., *Applying Sequential Analytic Methods to Self-Reported Information to Anticipate Care Needs*. EGEMS (Wash DC), 2016. 4(1): p. 1258.
18 Beaulieu, N.D., et al., *Concentration of high-cost patients in hospitals and markets*. Am J Manag Care, 2017. 23(4): p. 233-238.

19 Boscardin, C.K., et al., *Predicting cost of care using self-reported health status data*. BMC health services research, 2015. 15: p. 406.
20 Buck, J.A., J.L. Teich, and K. Miller, *Use of mental health and substance abuse services among high-cost Medicaid enrollees*. Adm Policy Ment Health, 2003. 31(1): p. 3-14.
21 Bynum, J.P.W., et al., *High-Cost Dual Eligibles' Service Use Demonstrates The Need For Supportive And Palliative Models Of Care*. Health Aff (Millwood), 2017. 36(7): p. 1309-1317.
22 Chang, H.Y., et al., *Identifying Consistent High-cost Users in a Health Plan: Comparison of Alternative Prediction Models*. Med Care, 2016. 54(9): p. 852-9.
23 Charlson, M.E., et al., *Medicaid managed care: how to target efforts to reduce costs*. BMC health services research, 2014. 14: p. 461.
24 Charlson, M., et al., *The Charlson comorbidity index can be used prospectively to identify patients who will incur high future costs*. PLoS One, 2014. 9(12): p. e112479.
25 Chechulin, Y., et al., *Predicting patients with high risk of becoming high-cost healthcare users in Ontario (Canada)*. Healthc Policy, 2014. 9(3): p. 68-79.
26 Cohen, S.B., T. Ezzati-Rice, and W. Yu, *The utility of extended longitudinal profiles in predicting future health care expenditures*. Med Care, 2006. 44(5 Suppl): p. I45-53.
27 Coughlin, T.A., T.A. Waidmann, and L. Phadera, *Among dual eligibles, identifying the highest-cost individuals could help in crafting more targeted and effective responses*. Health Aff (Millwood), 2012. 31(5): p. 1083-91.
28 Coughlin, T.A. and S.K. Long, *Health care spending and service use among high-cost Medicaid beneficiaries, 2002-2004*. Inquiry, 2009. 46(4): p. 405-17.
29 Crawford, A.G., et al., *Comparative effectiveness of total population versus disease-specific neural network models in predicting medical costs*. Dis Manag, 2005. 8(5): p. 277-87.
30 DeLia, D., Mortality, *Disenrollment, and Spending Persistence in Medicaid and CHIP*. Med Care, 2017. 55(3): p. 220-228.
31 de Oliveira, C., et al., *Patients With High Mental Health Costs Incur Over 30 Percent More Costs Than Other High-Cost Patients*. Health Aff (Millwood), 2016. 35(1): p. 36-43.
32 Figueroa, J.F., et al., *Characteristics and spending patterns of high cost, non-elderly adults in Massachusetts*. Healthc (Amst), 2017.
33 Figueroa, J.F., et al., *Concentration of Potentially Preventable Spending Among High-Cost Medicare Subpopulations: An Observational Study*. Ann Intern Med, 2017.
34 Fitzpatrick, T., et al., *Looking Beyond Income and Education: Socioeconomic Status Gradients Among Future High-Cost Users of Health Care*. Am J Prev Med, 2015. 49(2): p. 161-71.
35 Fleishman, J.A. and J.W. Cohen, *Using information on clinical conditions to predict high-cost patients*. Health Serv Res, 2010. 45(2): p. 532-52.
36 Ganguli, I., R. Thompson, and T.G. Ferris, *What can five high cost patients teach us about health care spending?* Journal of General Internal Medicine, 2016. 1): p. S469.
37 Graven, P.F., et al., *Preventable acute care spending for high-cost patients across payer types*. Journal of Health Care Finance, 2016. 42(3).
38 Guilcher, S.J., et al., *Who Are the High-Cost Users? A Method for Person-Centred Attribution of Health Care Spending*. PLoS One, 2016. 11(3): p. e0149179.

39 Guo, J.J., et al., *Characteristics and risk factors associated with high-cost medicaid recipients*. Manag Care Interface, 2004. 17(10): p. 20-27.

40 Hartmann, J., et al., *Analysing predictors for future high-cost patients using German SHI data to identify starting points for prevention*. Eur J Public Health, 2016. 26(4): p. 549-55.

41 Hensel, J.M., et al., *Rates of Mental Illness and Addiction among High-Cost Users of Medical Services in Ontario*. Can J Psychiatry, 2016. 61(6): p. 358-66.

42 Hirth, R.A., et al., *New Evidence on the Persistence of Health Spending*. Medical Care Research and Review, 2015. 72(3): p. 277-297.

43 Hunter, G., et al., *Health Care Utilization Patterns Among High-Cost VA Patients With Mental Health Conditions*. Psychiatr Serv, 2015. 66(9): p. 952-8.

44 Hwang, W., et al., *Persistent high utilization in a privately insured population*. Am J Manag Care, 2015. 21(4): p. 309-16.

45 Izad Shenas, S.A., et al., *Identifying high-cost patients using data mining techniques and a small set of non-trivial attributes*. Comput Biol Med, 2014. 53: p. 9-18.

46 Joynt, K.E., et al., *Segmenting high-cost Medicare patients into potentially actionable cohorts*. Healthc (Amst), 2017. 5(1-2): p. 62-67.

47 Joynt, K.E., et al., *Contribution of preventable acute care spending to total spending for high-cost Medicare patients*. JAMA, 2013. 309(24): p. 2572-8.

48 Krause, T.M., et al., *Future expenditure risk of silent members: a statistical analysis*. BMC Health Serv Res, 2016. 16: p. 319.

49 Ku, L.J., M.J. Chiou, and L.F. Liu, *Variations in the persistence of health expenditures and the implications for the design of capitation payments in Taiwan*. J Health Serv Res Policy, 2015. 20(3): p. 146-53.

50 Lauffenburger, J.C., et al., *Longitudinal Patterns of Spending Enhance the Ability to Predict Costly Patients: A Novel Approach to Identify Patients for Cost Containment*. Med Care, 2017. 55(1): p. 64-73.

51 Lee, N.S., et al., *High-Cost Patients: Hot-Spotters Don't Explain the Half of It*. J Gen Intern Med, 2017. 32(1): p. 28-34.

52 Leininger, L.J., et al., *Predicting high-need cases among new Medicaid enrollees*. The American journal of managed care, 2014. 20(9): p. e399-e407.

53 Lieberman, S.M., et al., *Reducing the growth of Medicare spending: geographic versus patient-based strategies*. Health Aff (Millwood), 2003. Suppl Web Exclusives: p. W3-603-13.

54 Meenan, R.T., et al., *Using risk-adjustment models to identify high-cost risks*. Med Care, 2003. 41(11): p. 1301-12.

55 Monheit, A.C., *Persistence in health expenditures in the short run: prevalence and consequences*. Med Care, 2003. 41(7 Suppl): p. III53-III64.

56 Pritchard, D., et al., *What Contributes Most to High Health Care Costs? Health Care Spending in High Resource Patients*. J Manag Care Spec Pharm, 2016. 22(2): p. 102-9.

57 Rais, S., et al., *High-cost users of Ontario's healthcare services*. Healthc Policy, 2013. 9(1): p. 44-51.

58 Reid, R., et al., *Conspicuous consumption: characterizing high users of physician services in one Canadian province*. J Health Serv Res Policy, 2003. 8(4): p. 215-24.

59 Reschovsky, J.D., et al., *Following the money: factors associated with the cost of treating high-cost Medicare beneficiaries*. Health Serv Res, 2011. 46(4): p. 997-1021.
60 Riley, G.F., *Long-term trends in the concentration of Medicare spending*. Health Aff (Millwood), 2007. 26(3): p. 808-16.
61 Robst, J., *Developing Models to Predict Persistent High-Cost Cases in Florida Medicaid*. Popul Health Manag, 2015. 18(6): p. 467-76.
62 Rosella, L.C., et al., *High-cost health care users in Ontario, Canada: demographic, socio-economic, and health status characteristics*. BMC Health Serv Res, 2014. 14: p. 532.
63 Snider, J.T., et al., *Identifying patients at risk for high medical costs and good candidates for obesity intervention*. Am J Health Promot, 2014. 28(4): p. 218-27.
64 Tamang, S., et al., *Predicting patient 'cost blooms' in Denmark: a longitudinal population-based study*. BMJ Open, 2017. 7(1): p. e011580.
65 Wammes, J.J.G., et al., *Characteristics and healthcare utilisation patterns of high-cost beneficiaries in the Netherlands: a cross-sectional claims database study*. BMJ Open, 2017. 7(11): p. e017775.
66 Wodchis, W.P., P.C. Austin, and D.A. Henry, *A 3-year study of high-cost users of health care*. Cmaj, 2016. 188(3): p. 182-8.
67 Zhao, Y., et al., *Identifying future high-cost cases through predictive modeling*. Disease Management and Health Outcomes, 2003. 11(6): p. 389-397.
68 Zulman, D.M., et al., *Multimorbidity and healthcare utilisation among high-cost patients in the US Veterans Affairs Health Care System*. BMJ Open, 2015. 5(4): p. e007771.
69 Bleich, S.N., et al., *Systematic Review of Programs Treating High-Need and High-Cost People With Multiple Chronic Diseases or Disabilities in the United States, 2008-2014*. Prev Chronic Dis, 2015. 12: p. E197.
70 Vuik, S.I., E.K. Mayer, and A. Darzi, *Patient Segmentation Analysis Offers Significant Benefits For Integrated Care And Support*. Health Aff (Millwood), 2016. 35(5): p. 769-75.
71 Hayes, S.L., et al., *High-Need, High-Cost Patients: Who Are They and How Do They Use Health Care? A Population-Based Comparison of Demographics, Health Care Use, and Expenditures*. Issue Brief (Commonw Fund), 2016. 26: p. 1-14.
72 Heider, D., et al., *Health care costs in the elderly in Germany: an analysis applying Andersen's behavioral model of health care utilization*. BMC Health Serv Res, 2014. 14: p. 71.
73 Wennberg, J.E., *Tracking Medicine:A Researcher's Quest to Understand Health Care: A Researcher's Quest to Understand Health Care*. 2010: Oxford University Press, USA.
74 Brinkman-Stoppelenburg, A., J.A. Rietjens, and A. van der Heide, *The effects of advance care planning on end-of-life care: a systematic review*. Palliat Med, 2014. 28(8): p. 1000-25.
75 Dixon, J., T. Matosevic, and M. Knapp, *The economic evidence for advance care planning: Systematic review of evidence*. Palliat Med, 2015. 29(10): p. 869-84.
76 Klingler, C., J. in der Schmitten, and G. Marckmann, *Does facilitated Advance Care Planning reduce the costs of care near the end of life? Systematic review and ethical considerations*. Palliat Med, 2016. 30(5): p. 423-33.
77 World Health Organization, *Strengthening of palliative care as a component of integrated treatment throughout the life course*. Journal of pain & palliative care pharmacotherapy, 2014. 28(2): p. 130-4.

78 Stadhouders, N., et al., *Policy options to contain healthcare costs: a review and classification*. Health Policy, 2016. 120(5): p. 486-94.
79 Miller, D.C., et al., *Large variations in Medicare payments for surgery highlight savings potential from bundled payment programs*. Health Aff (Millwood), 2011. 30(11): p. 2107-15.
80 Camacho, E.M., et al., *Long-term cost-effectiveness of collaborative care (vs usual care) for people with depression and comorbid diabetes or cardiovascular disease: a Markov model informed by the COINCIDE randomised controlled trial*. BMJ Open, 2016. 6(10): p. e012514.
81 Druss, B.G. and E.R. Walker, *Mental disorders and medical comorbidity*, in *Research Synthesis Report 21*. 2011, Robert Wood Johnson Foundation: Princeton, NJ.
82 Bodenheimer, T., *Strategies to Reduce Costs and Improve Care for High-Utilizing Medicaid Patients: Reflections on Pioneering Programs*. 2013, Centre for Health Care Strategies.
83 Tricco, A.C., et al., *Effectiveness of quality improvement strategies for coordination of care to reduce use of health care services: a systematic review and meta-analysis*. CMAJ, 2014. 186(15): p. E568-78.
84 Vrijhoef, B. and R. Thorlby, *Developing care for a changing population: supporting patients with costly, complex needs*. 2016, Nuffield Trust.
85 Sherry, M., et al., *Bridging the Silos of Service Delivery for High-Need, High-Cost Individuals*. Popul Health Manag, 2016.

Appendix 1. Final search strategy

Pubmed (restricted to Dutch, English, French and German):
(((((((High-cost*)) OR (high spending)) OR (Costliest)) OR (highest-cost*))) AND ((((((Patient*)) OR (Individual*)) OR (Benefici*)) OR (Person*)) OR (user*)) NOT medline[sb])) OR (((((("Economics, Hospital"[Mesh]) OR "Economics, Medical"[Mesh]) OR "Health Care Sector"[Mesh])) OR ("Costs and Cost Analysis"[Mesh]))) AND (((((((High-cost*)) OR (high spending)) OR (Costliest)) OR (highest-cost*))) AND ((((((Patient*)) OR (Individual*)) OR (Benefici*)) OR (Person*)) OR (user*))))

Embase:

1 (high-cost* or high spending or Costliest or highest-cost*).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
2 (patient* or individual* or benefici* or person* or user*).mp. [mp=title, abstract, subject headings, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword]
3 "hospital cost"/ or "health care cost"/ or "cost"/ or economic aspect/ or "hospital utilization"/ or medicare/ or exp medicaid/
4 1 and 2 and 3
5 ((high-cost* or high spending or Costliest or highest-cost*) adj3 (patient* or individual* or benefici* or person* or user*)).mp.
6 4 or 5

Appendix 2. Supplementary information

Author(s), country	Key points of the article
Aldridge and Kelly, United States	The majority of decedents were in the high-cost group, however the majority of high-cost patients were not in their last year of life. Not only is this group small (11%), the window of time for a significant impact on costs is limited by the patients' life expectancy. Findings confirm the need to focus on those with serious chronic illnesses, functional debility, and persistently high costs.
Ash et al., United States	Diagnosis-based risk models are at least as powerful as prior cost for identifying people who will be expensive. Combined cost and diagnostic data were even more powerful and more operationally useful, especially because the diagnostic information identifies the medical problems that may be managed to achieve better outcomes and lower costs.
Bayliss et al., United States	Self-reported health status, functional limitations, medication use, presence of 0-4 chronic conditions, self-reported ED use during the prior year, lack of prior insurance, age, gender, and deductible-based insurance product were predictive for high costs.
Beaulieu et al., United States	High-cost patients are only modestly concentrated in specific hospitals and markets. The hospitals and markets that disproportionately care for high-cost beneficiaries were markedly different than those that cared for fewer such patients: these hospitals were either academic teaching or for-profit institutions operating in urban settings and serve a greater proportion of low-income patients. Concentrated markets had a greater supply of specialists and a lower supply of long-term care beds. Spending in the last 6 months of life was also significantly higher in high-cost concentration HRRs.
Boscardin et al., United States	In addition to demographic characteristics and health service use, self-report of the presence of specific health conditions were predictive for high costs.
Buck et al., United States	Mental health/substance abuse service users constitute 11% of all Medicaid enrollees, but make up nearly a third of high-cost enrollees. Their use of non-mental health/substance abuse services is more important than their use of MH/SA services in determining their high-cost status. Adults account for two third of this high-cost MH/SA group, and they most frequently qualify for Medicaid through disability-related eligibility categories.
Bynum et al., United States	High combined Medicare and Medicaid spending are found in two distinct groups of high-cost dual eligibles: older beneficiaries who are nearing their end of life, and younger beneficiaries with sustained need for functional supports. High-cost dual eligibles often use costly inpatient settings, including acute care hospitals and inpatient long-term care services, in addition to nursing homes. 57% of high-cost dual eligibles reside in the community, not in long term care.
Chang et al., United States	Consistent high-cost users had higher total and pharmacy costs, and more chronic and psychosocial conditions than episodic high-cost users.
Charlson et al., United States	The comorbidity index was significantly correlated with the top 5% and top 10% of costs for the pooled sample, as well as for adults and children separately. Comorbidity can be used to identify beneficiaries most likely to incur high costs.
Charlson et al., United States	Prior year costs, prior year comorbidity, prior year DCG, and prior year hospitalizations were all evaluated as predictors of upper 5% and upper 10% of subsequent (2010) costs in separate models controlling for age, gender and mental health diagnosis. In adults, the comorbidity index was equivalent to DCG and prior cost in predicting the top 5% and 10% of cost, while prior hospitalization had much lower ability to identify such patients.
Chechulin et al., Canada	Age was a strong predictor of high costs, and as the material and social deprivation index increases, the risk of becoming high-cost increased. Males were more likely to incur high costs, and degree of rurality was also linked to high costs. Current and past healthcare utilization were the strongest predictors for high use. Several influential were significantly associated with high costs.

Cohen et al., United States	Prior year expenditures, frequency of prescribed medication purchases, the number of office based provider visits, activity limitations and health status were the most significant predictors for high costs. Other measures that were significantly related to high costs were age, gender, marital status, family income, living alone, and the presence of an infectious or respiratory condition. Predictive capacity of models did not suffer when restricted to a single year of prior information.
Coughlin et al., United States	20% of dual eligibles account for more than 60% of combined Medicaid and Medicare spending on the dual population. Subgroups were found among these high-cost population. Fewer than 1% of dual eligibles were in high-cost categories for both Medicare and Medicaid. Dual eligibles are a highly diverse group in terms of their spending. Being a dual eligible is not necessarily synonymous with high spending.
Coughlin and Long, United States	A high degree of spending persistence was observed: 57.9% of those in the top-10% remained in the top-10% in the two subsequent years. Two distinct high-cost groups were identified, those with persistently high costs and those with episodically high costs, each with different services driving their costs.
Crawford et al., United States	The following predictive factors, listed in descending order according to the magnitude of their importance statistics, were related to high costs: total medical costs, physician costs, prescription drug costs, number of unique diagnoses, age, number of prescription drug claims, number of unique procedures, hypertension symptoms, CAD symptoms, inpatient costs, and diabetes symptoms.
DeLia, United States	One forth of extreme spenders remained in that category in the three subsequent years. Almost all were blind, disabled and aged, the majority have a developmental disability, central nervous system diagnosis, or psychiatric diagnosis. Persistently high spenders were also more likely to be men, >40 years old, living in a nursing facility, or having a higher CDPS score.
de Oliveira et al., Canada	Mental health high-cost patients incurred 30% higher costs than other high-cost populations. They were younger, lived in poorer neighboorhouds, and had different health care utilization patterns.
Figueroa et al., United States	Characteristics and likelihood of high costs vary by major type of insurance. Nearly 1 in 5 Mediciad insured patients was likely to be high-cost (top-10%), these patients were more likely to be medically complex, with more chronic diseases and mental health health/substance abuse problems. Additionally, patterns of spending varied by major type of insurance.
Figueroa et al., United States	About 5% of total health care spending incurred by Medicare beneficiaries was potentially preventable, and most of this spending was incurred by high-cost patients. Large variations existed across high-cost subgroups. The high-cost frail elderly group accounted for nearly half of all potentially preventable spending after admissions for ACSCs or potentially avoidable ED visits. This spending was particularly high for heart failure, pneumonia, chronic obstructive pulmonary disease or asthma, and urinary tract infections.
Fitzpatrick et al., Canada	Future high costs status was most strongly associated with food insecurity, personal income, and non-homeownership. Living in highly deprived or low ethnic concentration neighborhoods also increased the odds of becoming an HCU.
Fleishmann et al., United States	Medical condition information substantially improved prediction of high expenditures beyond gender and age, with the DCG risk score providing the greatest improvement in prediction. The count of chronic conditions, self-reported health status, and functional limitations were significantly associated with future high expenditures, controlling for DCG score.
Ganguli et al., United States	Complex medical issues, physical disability/frailty, and mental illness/substance was linked with increased costs, while socioeconomic status, social network, activation, and trust in clinicians and the health system appeared to increase or decrease costs depending on context. Trust seemed to modify the interaction between patient activation and cost.

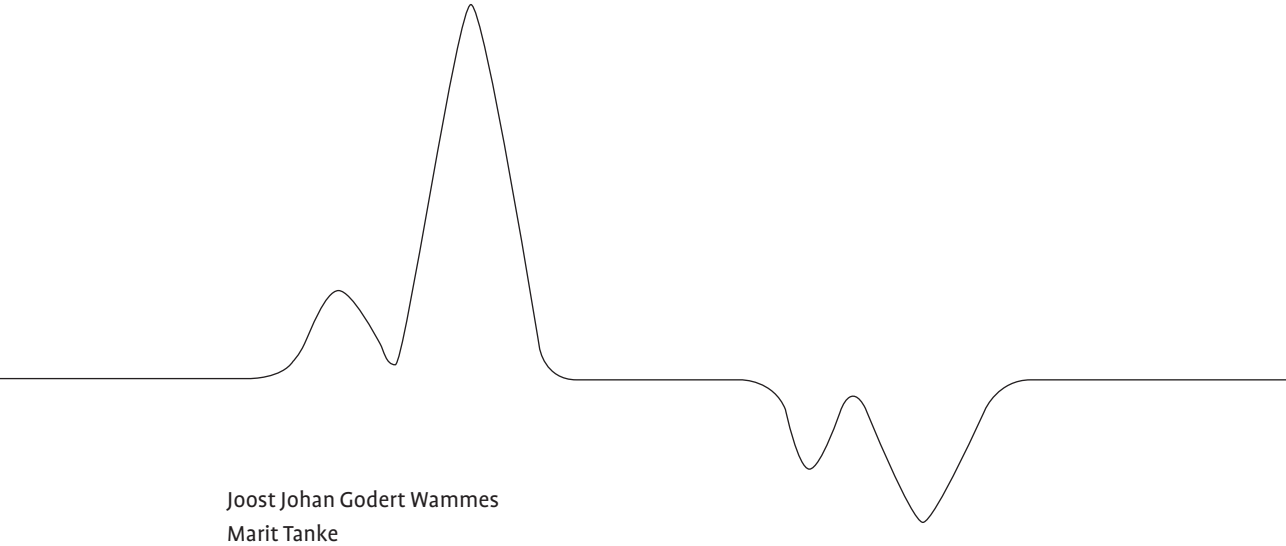
Graven et al., United States	Among the top-10%, 5.6%, 1.9%, and 3.8% was attributable to spending on preventable services for Medicaid, commercial, and Medicare Advantage patients, respectively. In the third year of spending among persistently high-cost patients in Medicaid, commercial and Medicaid advantage programs, cost were decreased by 11%, 25.6% and 30.6% respectively.
Guilcher et al., Canada	This study provides a novel methodological approach to categorize high-cost health system users into meaningful person-centered episodes. The most common clinical grouping categories to start a person-centered episode of care were Planned Surgical, Unplanned Medical and Post-Acute Admission Events. Inpatient acute and inpatient rehabilitation accounted for the largest proportions of costs.
Guo et al., United States	High-cost patients not only utilized more costly services, and more units of service per recipient, but also had higher per-unit costs for each of the service categories. The following groups had the highest odds of being a high-cost users: dying, disabled, urban resident, and male.
Hartmann et al., Germany	Several predictors were related to high costs, including insurance status (dependent coverage in particular), prior expenditures, home nursing, chronic diseases and multimorbidity, mental and behavioral disorders, musculoskeletal disorders, respiratory system disorders, cardiovascular diseases, and metabolic diseases.
Hensel et al., Canada	Seventeen percent of the most costly users had a prior diagnosis of a psychotic, major mood, or substance use disorder, and nearly 40% when anxiety and other disorders were included. The rate of mental illness and addiction rose incrementally across increasing user cost categories.
Hirth et al., United States	Individuals' positions within the spending distribution vary over time, but considerable persistence exists, particularly clear at the lower end of the spending distribution, but also at the top persistence is considerable. Many characteristics retained predictive power for future spending, including age, gender and a variety of medical conditions.
Hunter et al., United States	Approximately half of high-cost patients had at least one psychiatric diagnosis, and of these 49% had two or more psychiatric diagnoses. Utilization and costs of mental health and medical-surgical care differed among various groups of high-cost patients with mental health conditions.
Hwang et al., United States	Persistent high users had higher overall disease burden due to multiple chronic conditions and incurred significantly higher expenses in medication and professional services.
Izad Shenas et al., United States	Data mining techniques, including neural networks and decision trees, were used to identify non-trivial attributes of high-cost patients. Identified attributes were overall health perception, age, history of blood cholesterol check, history of physical/ sensory/ mental limitations, and history of colonic prevention measures.
Joynt et al., United States	High-cost beneficiaries were segmented into clinically relevant groups, including frail elders, those with disabilities or ESRD under the age 65, beneficiaries with chronic illnesses, and those who were relatively healthy at baseline. Frail elders were most likely to incur high costs, nearly half of the frail beneficiaries incurred high costs, and they comprised 40% of the high-cost population. Overall patterns of spending were relatively similar across high-cost segments, with inpatient spending contributing the largest share in general.
Joynt et al., United States	Approximately 10% of the costs for high-cost Medicare patients were deemed potentially preventable. The percentage was slightly higher for the persistently high-cost cohort. Hospital referral regions with a higher primary care or physician supply had higher annual preventable costs per capita.
Krause et al., United States	Silent-members are members of a medical health plan who submit no claims for healthcare services in a benefit year despite 12 months of continuous-enrollment. This study found that silent members who seek care in subsequent years have a greater probability of becoming high-expenditure claimants than those with low-expenditure experience.
Ku et al., Taiwan	Of the top-10%, 39% remained high-cost in the year thereafter. NHI expenditure percentiles, and all chronic conditions significantly predicted future expenditures.

Lauffenburger et al., United States	High-cost patients had higher mean comorbidity scores (measured using four risk adjustment measures). Trajectory modeling may be a useful way to predict costly patients that could be implementable by payers to improve cost-containment efforts.
Lee et al., United States	Five distinct phenotypes of high-cost patients with diverse drivers of cost were identified. Besides, "hot-spotters" (those with four or more admissions) were quantified. They accounted for 9% of high-cost patients and 19% of that population's costs. The majority of "hot-spotters" were in the cluster of patients who had 'frequent care'.
Leininger et al., United States	Self reported health measures were meaningful predictors of high costs, this included individual conditions, behavioral variables, prescription drug use, previous year utilization, and access to care measures.
Lieberman et al., United States	This paper explored the potential of two alternative approaches for reducing the rate of growth in Medicare spending. Viewed from a budgetary perspective, concentration in Medicare spending suggests the importance of focusing on high-spending patients. Spending per beneficiary correlated strongly with inpatient use. The prevalence of serious chronic conditions is higher among high-spending beneficiaries. A high-cost patient was five times more likely to die. However, only one fifth died at the end of the year.
Meenan, United States	This study evaluated a variety of risk models to predict high-cost patients. To predict top-1% and top-0.5%, ACGs, DCGs, GRAM, and Prior-expense were very comparable in overall discrimination (AUCs, 0.83– 0.86). DCGs captured the most "high-cost" dollars among enrollees with asthma, diabetes, and depression; predictive performance among demographic groups (Medicaid members, members over 64, and children under 13) varied across models.
Monheit, United States	A sizeable minority of high expenditure cases exhibits persistently high expenditures in the short run. However, when all persons in a top expenditure percentile are considered, health expenditures do begin to regress to the mean over time as a majority of high spenders move to lower positions throughout the expenditure distribution.
Powers and Chaguturu, United States	Little is known about variation in clinical characteristics and care-utilization patterns among payer-defined groups. The costliest 1% of Medicare patients had an average of 8 co-occurring chronic conditions. In Medicaid, high-cost patients also had several co-occurring chronic conditions (five on average) but there was a striking prevalence of mental health disorders. In commercial populations, high-cost patients had fewer chronic conditions and were more likely to have disease risk factors than end-stage sequelae. Drivers of high costs in this population included catastrophic injuries, neurologic events, and need for specialty pharmaceuticals.
Pritchard et al., United States	Spending pattern for high-cost patients differs considerably from the general population. The absolute expenditures for each place of service were increased, and the share of spending on inpatient services is significantly higher in high-cost patients, while the share of expenditures attributed to major outpatient places of service and pharmacy are lower. Common health conditions, such as back disorders and osteoarthritis, contribute a large share of expenditures, but other conditions such as chronic renal failure, graft rejection, and some cancers accounted for disproportionately higher expenditures in high-cost patients.
Rais et al., Canada	Males are more costly than females. Seniors accounted for the majority of high-cost users and costs, but the average costs per patients decreased with age. Of the different clinical conditions, circulatory system conditions incurred the most costs.
Reid et al., Canada	High-cost users are overwhelmingly characterized by multiple and complex health problems. This relatively small group accounted for a disproportionate share of primary care and specialist encounters as well as inpatient days.

Reschovsky et al., United States	Among high-cost patients, health was the predominant predictor of costs, with most physician and practice and many market factors (including provider supply) insignificant or only weakly associated with high costs. Beneficiaries whose usual physician was a medical specialist or reported inadequate office visit time, medical specialist supply, provider for-profit status, care fragmentation, and Medicare fees were associated with higher costs.
Riley , United States	Annual expenditures became less concentrated over time, although the year-to-year persistence of person-level high costs remained strong. There was an increase in the prevalence of chronic conditions among high-cost beneficiaries. Spending concentration in Medicare decreased over time, perhaps due to 1) trends in longevity and medical expenses (increasing life expectancy has had the effect of spreading the same level of healthcare costs over a greater number of years; as age of death increases, lifetime Medicare costs increase only slightly), 2) expensive technologies are increasingly used on less sick patients, or 3) trends in disability.
Robst, United States	High costs were very persistent, as a high percentage of individuals were high-cost cases for multiple years. In addition, individuals receiving ICF-mental retardation services were very likely to have persistent high costs. Individuals with 1 or more inpatient stays in the base year were less likely to remain high cost in the future. Most high-cost cases had multiple diagnoses.
Rosella et al., Canada	High-cost patients tended to be older with multiple comorbidities and were more likely to be white, female and have lower household income. Risky behaviors were not overwhelmingly drivers of short term high-cost, but this is likely an artifact.
Snider et al., United States	A logistic model was used to capture the effect of BMI on the risk of high future medical spending. Individuals in all obesity classes have higher risk of high medical spending in the following year compared to normal weight patients (BMI ≤ 25).
Tamang et al., Denmark	Cost bloomers (those who move from the lower to the upper percentile in one year) represented the majority of high-cost patients. They were younger, had less comorbidity, lower mortality and fewer chronic conditions. Diverse population health data, in conjunction with modern statistical learning methods for analyzing large data sets, can improve prediction of future high-cost patients over standard diagnosis-based tools, especially for cost-bloom prediction task.
Wammes et al., Netherlands	Expensive treatments, most cost-incurring condition and age proved to be informative variables for studying high-cost patients. Expensive care use (expensive drugs, ICU treatment, dialysis, transplant care and DRG >€30 000) contributed to high costs in one third of top 1% beneficiaries and in less than 10% of top 2%–5% beneficiaries. High-cost beneficiaries were overwhelmingly treated for diseases of circulatory system, neoplasms and mental disorders. More than 50% of high-cost beneficiaries were 65 years of age or younger, and average costs decreased sharply with higher age within the top 1% population.
Wodchis et al., Canada	High health care costs were related to a diverse set of patient health care needs and were incurred in a wide array of healthcare settings. Analyses showed moderate stability in health care costs for individuals over a 3-year period. High-cost spending patterns and conditions varied across age groups.
Zhao et al., United States	This study evaluated three models to predict high-cost patients, including a DCG-model, a prior cost model, and a prior plus DCG-model (combo model). The DCG-model and combo model outperformed the prior cost model.
Zulman et al., United States	Multisystem morbidity is common in high-cost patients, approximately two-thirds have chronic conditions affecting three or more body systems. While some patients with cancer or mental illness may benefit from disease specific interventions, the majority most likely require programs that address their heterogeneous health needs.

CHAPTER 7

Characteristics and healthcare utilization patterns of high-cost beneficiaries in the Netherlands; a cross-sectional claim database study



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Abstract

Objective To determine medical needs, demographic characteristics and healthcare utilization patterns of the top-1% and top-2-5% high-cost beneficiaries in the Netherlands.

Design Cross-sectional study using 1-year claim data. We broke down high-cost beneficiaries by demographics, the most cost-incurring condition per beneficiary, and expensive treatment use.

Setting Dutch curative health system, a health system with universal coverage.
Participants: 4.5 million beneficiaries of one health insurer.

Measures annual total costs through hospital, intensive care unit use, expensive drugs, other pharmaceuticals, mental care, and others; demographics; most cost-incurring and secondary conditions; inpatient stay; number of morbidities; costs per ICD10-chapter; and expensive treatment use (including dialysis, transplant surgery, expensive drugs, intensive care unit and DRGs > €30,000).

Results The top-1% and top-2-5% beneficiaries accounted for 23% and 26% of total expenditures respectively. Among top-1% beneficiaries, hospital care represented 76% of spending, of which respectively 9.0% and 9.1% were spent on expensive drugs and ICU care. We found that 54% of top-1% beneficiaries were aged 65 or younger, and that average costs sharply decreased with higher age within the top-1% group. Expensive treatments contributed to high costs in one third of top-1% beneficiaries, and in less than 10% of top-2-5% beneficiaries. The average number of conditions was 5.5 and 4.0 for top-1% and top-2-5% beneficiaries respectively. 53% of top-1% beneficiaries were treated for circulatory disorders, but for only 22% of top-1% beneficiaries this was their most cost-incurring condition.

Conclusions Expensive treatments, most cost-incurring condition, and age proved to be informative variables for studying this heterogeneous population. Expensive treatments play a substantial role in high-costs beneficiaries. Interventions need to be aimed at beneficiaries of all ages; a sole focus on elderly would leave many high-cost beneficiaries unaddressed. Tailored interventions are needed to meet the needs of high-cost beneficiaries, and to avoid waste of scarce resources.

Strengths and limitations of this study

- This study presents an in-depth analysis of the medical needs, demographics and healthcare utilization of high-cost beneficiaries in the Netherlands.
- We characterized high-cost beneficiaries and spending patterns using several variables, including expensive treatment use (e.g. dialysis, expensive drugs, ICU), most cost-incurring condition, and age.
- Analyses were limited to one large insurer, but its beneficiaries are representative for the Netherlands.

Introduction

It is known that health care costs are concentrated among small numbers of 'high-cost' beneficiaries. These high-cost beneficiaries are the sickest and most complex populations. Although they receive substantial care from multiple sources, critical health care needs are often unmet, and many receive unnecessary and ineffective care [1-4]. Therefore, high-cost beneficiaries are a useful group on which to focus efforts of quality improvement and cost containment.

For effective quality improvement and cost reduction it is necessary to acquire an in-depth understanding of the characteristics, health care use and other factors that drive the costs of these groups of high-cost beneficiaries [5,6]. Current literature suggests that a high prevalence of multiple (chronic) conditions may explain high-cost beneficiaries' excessive care use [7,8]. This presence of multimorbidity among high-cost beneficiaries makes them difficult to understand: how to characterize patients that suffer from several diseases? Lehnert et al [9] found that the number of chronic comorbidities were nearly exponentially related to costs: the higher the number of chronic comorbidities, the higher the costs of an additional comorbidity. Based on this study, we hypothesized that in high-cost beneficiaries the most cost-incurring condition accounts for a disproportionate share of costs, and that secondary conditions account for the remainder of costs.

A major limitation of current literature is that little is known about patterns in care use and characteristics among different age groups [10]. In addition, until today no studies have reported the role of expensive treatments (e.g. expensive drugs, transplant surgery, intensive care units, dialysis) as drivers of high costs. Further insight in healthcare utilization patterns is needed to develop interventions and inform policy aimed at high-need, high-cost populations.

The primary aim of this study was to determine medical needs, demographic characteristics and healthcare utilization patterns of high-cost beneficiaries in the Netherlands. We first determined characteristics and spending and quantified the share of high-cost beneficiaries that use expensive treatments. We then used a beneficiary's most cost-incurring medical condition to examine characteristics and utilization patterns. In addition, we compared utilization and conditions across age groups. All analyses were performed for top-1% and top-2-5% beneficiaries separately. This distinction is often used in literature [11-14] and may improve understanding of high-cost beneficiaries.

Methods

Design and context

We conducted a cross-sectional study using claims data from 2013 in the Netherlands. In the Netherlands, the Health Insurance Act legally requires health insurers to provide a nationally set benefits package. Nearly universal coverage for curative care is achieved through mandatory purchase of statutory private health insurance [15,16]. Analyses were done in-house with Zilveren Kruis, a health insurer covering 4.5 million beneficiaries who were primarily living in the central, eastern and western parts of the Netherlands. The basic

principle of the Dutch curative health system is that insurers compete for beneficiaries, and that they act as prudent buyers of services for their beneficiaries. Health insurers operate nationwide, are obliged to accept all applicants for basic health plans and are not permitted to risk-rate premiums for these basic plans. Every insured person, aged 18 years or older, is required to pay an annual deductible (350 euro in 2013), from which some services, such as general practice visits, are excluded. In addition to the basic health plan, more than 80% of the population buys voluntary insurance. Premiums for voluntary insurance are not regulated, and insurers are allowed to screen applicants. The system provides a wide range of services, including care provided by general practitioners, hospitals, and specialists; dental care through age 18; prescription drugs; physiotherapy through age 18; most mental care; medical aids and devices; maternity care; transportation and others. In our study we also included private voluntary supplementary insurance which typically covers dental care, some allied healthcare (including physiotherapy, occupational therapy, dietary advice, speech therapy) and alternative medicine (typically homoeopathy, acupuncture, natural medicine, magnetizing and osteopathy).

Data

All insured in 2013 were included in this study. Several beneficiary characteristics were obtained from the insurer's databases, including gender, socio-economic status based on income estimates per postal code, date of birth and date of death (until February 20th, 2015). Date of death was categorized to four quarters in 2013 and any date post-2013. More information about (a predecessor of) this database is provided in Smeets et al [17].

Total costs per beneficiary were calculated by summing all claims with a starting date in 2013. We defined the beneficiaries with the top-1% and the top-2-5% of total costs as two groups of high-cost beneficiaries. The remaining 95% were categorized as low-cost beneficiaries. All claims were categorized in nine cost groups (health sectors) using a link table provided by the Dutch Healthcare Institute. These sectors included: hospital care (including care used abroad), mental health care, primary care, maternal care, allied health care, outpatient pharmaceutical prescriptions, medical devices, dental care (most dental care is reimbursed through complementary insurance benefits), and voluntary complementary insurance benefits.

Below, we describe how we operationalised the variables that we included in our analysis, including the treatment costs per diagnosis, the prevalence of conditions and multimorbidity count, and the use of specific (expensive) services.

Treatment cost per diagnosis

We categorized and analyzed hospital and mental care costs, according to the ICD10 international classification of diseases [18]. Treatment costs were categorized to the level of ICD10-chapters (e.g. chapter IX: diseases of the circulatory system) and ICD10-subchapters (e.g. subchapter I60-I69 cerebrovascular diseases).

The great majority of hospital care in the Netherlands is reimbursed through payment products similar to Diagnosis Related Group (DRGs, which cover both in- and outpatient hospital care) and so-called add-ons for expensive drugs and treatment at the intensive care unit (ICU). To compute treatment costs per diagnosis, the DRGs were categorized using a

link table provided by the Dutch Health Care Authority. This link table (version 22 December 2014) was developed to categorize hospital claims to specific health care needs, following the ICD10 classification [18]. For the purpose of our study, we made a few minor corrections to the link table. As we found the ICD-subchapter I30-I52 (other circulatory diseases) highly prevalent but not informative, we decided to disaggregate this subchapter. Add-ons were not used for establishing treatment cost per diagnosis, but are dealt with separately (see "use of specific expensive services").

In 2013, the Dutch mental care sector consisted of 'primary' mental care, such as care provided at general practices, by psychologists and psychotherapists, and at 'secondary' or specialized mental care provided in mental care institutions. Only claims from secondary mental health care were used for characterization as these specify information about diagnoses and treatment. These claims were categorized to the ICD10-(sub)chapter and added to the hospital claims for ICD-10 chapter V: mental and behavioural disorders. Additionally, the number of inpatients days in mental care per beneficiary was calculated (but not used for establishing treatment cost).

Prevalence of conditions and multimorbidity count

Prevalence of conditions was established using the same categorization as described above. In addition, we used parameters from the Dutch risk-adjustment scheme: pharmaceutical cost groups that indicate chronic use of drugs for different conditions. These pharmaceutical cost groups were categorized to ICD10-(sub-)chapters and integrated with the former to establish prevalence of conditions. A detailed description of the Dutch risk-adjustment scheme is provided in van Veen et al [19]. Multimorbidity was operationalised in three ways. First, multimorbidity was calculated by counting the number of prevalent ICD10-chapters per beneficiary. Second, we counted the number of prevalent ICD10-subchapters per beneficiary. Third, the number of pharmaceutical cost groups was counted, reflecting the number of chronic multimorbidities.

Use of specific (expensive) services

We developed dummy variables for specific types of care. Beneficiaries were regarded 'expensive care users' if their claims included a minimum of €10,000 for 'add-ons' for ICU treatment or expensive drugs. We used €10,000 as threshold because in 2013 expensive drugs only qualified for add-on reimbursement when average yearly costs per beneficiary exceeded this value. ICU treatment as reimbursed through add-ons included ICU treatment days, ICU consultations, ICU surcharges for specific services, ICU neonatal and pediatric care, and ICU transport services such as inter-clinical transportation services and Mobile Intensive Care Unit (MICU). Expensive drugs reimbursed through add-ons included growth hormones, antineoplastic agents, TNF-alfa inhibitors, orphan drugs, haemostatics and other expensive drugs [20]. The list of drugs and indications that qualify for add-on reimbursement can be found at www.farmatec.nl.

A separate dummy variable 'transplant' was developed, for beneficiaries who received a transplant or transplant-related care (both pre- and post transplant). One DRG-description that included the word 'transplant' was sufficient for a person to qualify as transplant-beneficiary. Similarly, the variable 'dialysis' was created for all beneficiaries receiving dialysis

for renal failure (both peritoneal and hemodialysis). In addition, all DRGs with an average price > €30,000 were identified and together included as separate binary variable. This price was chosen as all top-1% beneficiaries incurred €30,000 or more. Furthermore, two dummy variables for mental health use were computed, the first on mental care use (> €0 mental care costs) and the second on inpatient stays (> 0 days). The total number of inpatient hospital days per beneficiary was estimated using national averages of hospital days per DRG [21]. Finally, we used claim data to derive the number of different hospitals, university medical centers, and hospital specialisms that beneficiaries were treated at, as well as the number of ambulance transportations, and emergency department visits. For full details concerning the variable computation, please contact the corresponding author.

Analyses

We explored the composition of expenditures across health sectors for both top-1% and top-2-5% beneficiaries. Demographics, medical characteristics and (expensive) health care use were analyzed using descriptive statistics.

Most cost-incurring and secondary conditions

For each high-cost beneficiary we identified the most cost-incurring ICD10-(sub)chapter. For both top-1% and top-2-5% beneficiaries, we first determined the prevalence of each ICD10-subchapter. Second, for both high-cost groups we summed treatment cost per ICD10-subchapter, and divided this with the sum of total costs. Third, for each ICD10-subchpater we calculated how frequently it was the most cost-incurring condition for the beneficiaries in these groups. Fourth, we divided the percentage of beneficiaries with a ICD10-subchapter as the most cost-incurring condition by the overall prevalence of the ICD10-subchapter. This metric was used to distinguish between ICD10-subchapters that were mainly found as most cost-incurring conditions compared to ICD10-subchapters that were mainly found as secondary conditions. Fifth, for each beneficiary we divided the treatment cost for the most cost-incurring condition by total costs. This figure was averaged for each ICD10-subchapter and determines the contribution of these conditions to total costs within the subpopulation.

Health care use according to most cost-incurring ICD10-chapter and across age groups

To identify patterns in (expensive) health care use, we developed cross-tables with costs per ICD10-chapter, (expensive) health care use indicators and demographic characteristics as descriptive variables. Beneficiaries were selected by the most cost-incurring ICD10-chapter, to prevent that beneficiaries with multimorbidity would be counted several times.

Finally, we compared utilization patterns and conditions across age groups. We examined total costs, spending per sector and we identified the five most cost-incurring ICD10-chapters per age group.

All analyses were performed using SAS 9.4, Enterprise Guide 6.1.

TABLE 1 General characteristics and indicators for health care use for three distinct cost groups.

General characteristics		Top-1%	Top-2-5%	Bottom-95%
Number of beneficiaries		45,207	180,826	4,294,611
Average total costs (SD)		€56,424 (€40,830)	€15,780 (€5,208)	€1,345 (€1,773)
Share of total costs		22.8%	25.5%	51.7%
Private spending† (SD)		€330 (€172)	€335 (€165)	€159 (€181)
Gender	Male:	52.3%	44.8%	49.6%
	Female:	47.7%	55.2%	50.4%
Mean age (SD)		58.5 (21.6)	58.0 (21.8)	39.2 (23.3)
Median age		64	62	39
Percentage dying in or after study period	Q1:	0.7%	0.9%	0.2%
	Q2	1.8%	1.6%	0.2%
	Q3	3.2%	1.7%	0.1%
	Q4	4.2%	1.9%	0.1%
	>Q4‡:	12.5%	5.9%	0.7%
Socioeconomic status >15 inhabitants††:		4.5%	3.1%	1.0%
Lowest incomes:		31.1%	31.5%	31.4%
Average income:		37.5%	38.5%	37.7%
High income:		26.8%	26.7%	28.6%

Medical characteristics	Top-1%	Top-2%-5%	Bottom-95%
Average number of comorbidities – ICD-chapter (SD)	4.2 (2.1)	3.3 (1.8)	0.7 (1.1)
Average number of comorbidities – ICD-subchapter (SD)	5.5 (3.1)	4.0 (2.3)	0.8 (1.2)
Average number of chronic comorbidities – calculated by pharmaceutical cost groups (SD)	1.1 (1.2)	1.0 (1.1)	0.2 (0.6)

(Expensive) healthcare use	Top-1%	Top-2%-5%	Bottom-95%
Percentage using expensive care > €10,000	24.6%	5.8%	0.0%
Percentage transplant beneficiaries	3.7%	0.8%	0.03%
Percentage receiving dialysis	6.1%	0.1%	0.0%
Percentage receiving DRG > €30,000	4.5%	0.03%	0%
Percentage with >0 inpatient mental health care stays	13.0%	3.3%	0.04%
Percentage with mental health care costs >€0	23.5%	20.6%	6.4%
Average number of inpatient mental hospital days‡‡	54.7 (74.3)	4.0 (11.7)	0.05 (0.8)
Percent visiting a specialized mental care center	22.5%	19.2%	4.8%
Average number of hospital specialists involved (SD)	4.2 (2.3)	3.0 (1.8)	0.6 (1.0)
Average number of hospitals visited (SD)	1.9 (1.0)	1.6 (0.9)	0.5 (0.7)
Average number of inpatient hospital days (SD)	22.3 (26.0)	7.2 (8.4)	0.4 (1.5)
Percentage using care at a university hospital	39.7%	25.8%	4.5%
Average number of ambulance transportations (SD)	1.4 (4.3)	0.5 (1.0)	0.02 (0.17)
Average number of emergency department visits (SD)	0.7 (1.4)	0.4 (0.7)	0.07 (0.27)

† Consisting of the compulsory deductible of €350. ‡ Dates of death were recorded until the 20th of February 2015.

†† Most of whom are institutionalized. ‡‡ For those with mental health care costs >€0.

Results

General breakdown of costs

Average total costs for top-1%, top-2-5% and bottom-95% beneficiaries were €56,424, €15,780 and €1,345 respectively, representing 22.8%, 25.5% and 51.7% of total spending (table 1). For top-1% beneficiaries, hospital care represented 76% of costs, of which respectively 9.0% and 9.1% were for expensive drugs and ICU care. 12.7% and 6.6% of costs were for mental health care and outpatient pharmaceuticals. For top-2-5% beneficiaries, hospital care represented 59.7% of spending, of which 6.0% and 2.1% were spent on expensive drugs and ICU care. 9.8% and 11.2% were spent on mental health care and outpatient pharmaceuticals.

Demographics and (expensive) healthcare use

Table 1 presents demographic and medical characteristics of the study population as well as (expensive) healthcare use. Males were overrepresented among top-1% beneficiaries, and females were overrepresented among top-2-5% beneficiaries. Top-1% and top-2-5% beneficiaries were much older than low-cost beneficiaries. Furthermore, high-cost beneficiaries were more likely to die: 9.9% and 6.1% of top-1% and top-2-5% beneficiaries died. However, 63.7% of beneficiaries in our study who died in 2013 or later did not incur high costs in 2013. The average number of morbidities based on ICD10-subchapters for top-1%, top-2-5% and bottom-95% beneficiaries was 5.5, 4.0 and 0.8 respectively.

Table 1 also shows that top-1% and top-2-5% beneficiaries scored higher than low-cost users for each specific service, and top-1% beneficiaries scored higher than top-2-5% beneficiaries. Both top-1% and top-2-5% beneficiaries used on average one type of drugs (pharmaceutical cost groups) continuously. 24.8% of top-1% and 5.8% of top-2-5% beneficiaries incurred more than €10,000 on expensive drugs and ICU. Furthermore, 6.1% of top-1% beneficiaries underwent dialysis and 3.7% received transplant care. Top-1% and top-2-5% beneficiaries were treated in on average 1.9 and 1.6 hospitals, and used on average 22 and 7 inpatient days respectively. Finally, 13% and 3.3% of top-1% and top-2-5% beneficiaries were admitted to mental care institutions, respectively.

Utilization according to ICD10-subchapters, and most cost-incurring and secondary conditions

Appendix 1 presents five parameters for both high-cost populations. Among those in the top-1%, a high prevalence of several cardiovascular diseases, COPD, diabetes mellitus, and depression were found. In addition, the total treatment costs for renal insufficiency (including dialysis) were much higher than for any other ICD10-subchapter, and accounted for 6.8% of total costs among top-1% beneficiaries. We use table 2, with a selection of ten ICD10-subchapters in top-1% beneficiaries, to illustrate the other parameters for top-1% beneficiaries. Renal insufficiency, certain cancers, and several cardiovascular diseases were frequently found as the most cost-incurring condition among top-1% beneficiaries. Furthermore, for beneficiaries that were treated for cancer, the cancer itself was in most cases the most cost-incurring condition (e.g. 74.3% of beneficiaries with leukemia). In contrast, circulatory disorders were mainly found as secondary condition: for example, in less than 30% of patients with ischemic heart disease or heart failure this was their most

cost-incurring condition. Finally, we determined the contribution of ICD10-subchapters towards total costs per beneficiary. The most cost-incurring condition accounted for 40-70% of total costs per beneficiary, depending on the ICD10-subchapter.

TABLE 2 Ten conditions with highest total costs among top-1% beneficiaries.

	Prevalence[a]	% of total costs[b]	% as most cost-incurring condition[c]	% most cost-incurring/ prevalence[d]	% of costs by most cost-incurring condition[e]
N17-N19 Renal failure	12.2%	6.8%	6.4%	52.4%	66.0%
C81-C96 Leukemia	5.6%	3.0%	4.1%	74.3%	41.4%
C15-C26 Malignant neoplasms of digestive organs	7.5%	2.4%	5.4%	71.2%	47.9%
I60-I69 Cerebrovascular diseases	7.9%	2.1%	4.2%	53.1%	52.7%
I70-I79 Diseases of arteries, arterioles and capillaries	9.6%	2.0%	4.1%	42.7%	47.3%
C30-C39 Lung cancer	5.9%	1.7%	3.5%	59.1%	52.5%
I51-I52 Complications/ ill-defined descriptions, other heart disorders	9.6%	1.6%	3.2%	33.1%	50.3%
I44-I49 Atrial fibrillation, rhythm and conduction disorders	11.8%	1.6%	2.9%	24.3%	58.5%
I20-I25 Ischemic heart diseases	12.7%	1.6%	3.7%	29.0%	41.9%
I50 Heart failure	9.3%	1.5%	2.6%	28.4%	57.1%

[a] Prevalence of each ICD10-subchapter among top-1% beneficiaries. E.g. 12.2% of top-1% beneficiaries were treated for renal failure.

[b] Sum of total treatment costs per ICD10-subchapter. E.g. treatment of renal failure accounted for 6.8% of total expenditures of top-1% beneficiaries.

[c] Percentage of top-1% with this ICD10-subchapter as most cost-incurring condition. E.g. 6.4% of top-1% beneficiaries had renal failure as most cost-incurring condition.

[d] Percentage most cost-incurring condition relative to prevalence: fourth column divided by second column. E.g. for 52.4% of top-1% beneficiaries who were treated for renal failure, this was also their most cost-incurring condition.

[e] Percentage of costs accounted for by the most cost-incurring condition. E.g. among top-1% beneficiaries with renal failure as most cost-incurring condition, this disease accounted for on average 66% of total costs per beneficiary.

Utilization according to most cost-incurring ICD10-chapter

Table 3 and appendix 2 show cross-tables for spending, demographics and indicators for (expensive) healthcare use. In these analyses, beneficiaries were selected by most cost-incurring ICD10-chapter, to avoid multimorbid beneficiaries being analyzed on multiple

rows. Among top-1% beneficiaries, three ICD10-chapters were frequently found as most cost-incurring ICD10-chapter: mental or behavioral disorders, neoplasms and diseases of the circulatory system. These groups had quite different characteristics and utilization. Beneficiaries with mental or behavioral disorders were relatively young, had a low number of morbidities and low mortality. Beneficiaries with neoplasms were the largest subgroup with high mortality. Beneficiaries with diseases of circulatory system were oldest (on average 69 years old) and predominantly men. Expensive drugs were heavily concentrated among beneficiaries with neoplasms. ICU costs were distributed more proportionally; a quarter was incurred by beneficiaries with circulatory diseases.

Among top-2-5% beneficiaries, the same three most cost-incurring ICD10-chapters predominated, albeit they represented a smaller share of the group. Several other ICD10-chapters had relatively high costs, including diseases of the digestive system; injury, poisoning and certain other consequences of external causes (femur fracture most prominently); and diseases of the musculoskeletal system and connective tissue. Beneficiaries with neoplasms; diseases of the respiratory system; and symptoms, signs and abnormal clinical and laboratory findings most frequently died. Expensive drugs were primarily used by beneficiaries with diseases of the musculoskeletal system (rheumatoid arthritis), neoplasms and diseases of the digestive system.

Health care use across age groups

Figure 1 and appendix 3 provide an overview of cost segments per age category among top-1% and top-2-5% beneficiaries. With the exception of infants, treatment at the ICU represented a maximum of 10% of costs per age group. Moreover, treatment at the ICU represented a major cost driver primarily among top-1% beneficiaries. The proportion of costs spent on expensive drugs was highest (13.4% of total costs) among top-1% beneficiaries between 21 and 30 years old. Mental care accounted for a large share of costs among children and young and middle aged adults. The percentage of cost incurred by outpatient and non-expensive pharmaceuticals was more pronounced among top-2-5% beneficiaries than among top-1% beneficiaries.

Table 4 and appendix 4 present the five ICD10-chapters with highest total costs per age group for top-1% and top-2-5% beneficiaries. As mentioned before, we found that high-cost beneficiaries are generally older than low-cost beneficiaries. However, table 4 shows that within the top-1% beneficiaries average costs decreased with higher age: average costs ranged from €47,000 on average for top-1% beneficiaries over 80 of age to >€80,000 on average for infants. In addition, 54% of top-1% and 57% of top-2-5% beneficiaries were 65 years of age or younger.

For each age group, there were different ICD10-chapters with highest costs. Among top-1% beneficiaries, cardiovascular diseases and diseases of the genitourinary system gained importance with higher age, whereas mental and behavioral disorders predominated among younger and middle-aged beneficiaries. Among top-2-5% beneficiaries, a similar pattern of diseases across age groups was observed. However, pregnancy-related conditions played a more significant role among beneficiaries between 20 and 40 years of age, and musculoskeletal conditions played a more significant role in several age groups than they did among top-1% beneficiaries.

TABLE 3 Cross table describing patterns of health care use and demographics of top-1% beneficiaries.

ICD10-chapter	Prevalence[a]	% most cost-incurring ICD10-chapter	Average age	Percentage dying	Percentage men	Average number of comor (ICD-chapter)	Average cost (*€1000)	Expensive drugs (*€1000)	Intensive care (*€1000)	Neoplasm (*€1000)	Mental and behavioural disorders (*€1000)	Diseases circulatory system (*€1000)	Diseases genitourinary system (*€1000)
Certain infectious and parasitic diseases	10.2%	1.0%	60.3	16.8%	60.4%	5.5	54	2,515	3,407	407	247	382	400
Neoplasms	36.8%	23.2%	62.0	16.4%	49.5%	3.9	56	111,927	27,734	295,902	2,062	7,629	4,991
Diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism	4.4%	1.0%	42.1	3.7%	65.9%	3.3	103	33,535	531	143	62	213	91
Endocrine, nutritional and metabolic diseases	21.5%	1.3%	39.2	6.1%	49.2%	4.0	67	8,452	3,025	191	228	432	224
Mental and behavioural disorders	32.2%	14.7%	41.1	1.2%	51.8%	2.7	55	4,487	4,292	841	135,431	1,746	787
Diseases of the nervous system	29.3%	2.6%	54.7	8.4%	52.2%	4.6	56	9,303	6,185	845	682	1,064	442
Diseases of the eye and adnexa	18.2%	0.2%	62.4	1.1%	43.6%	4.0	47	974	112	45	15	13	9
Diseases of the ear and mastoid process	5.4%	0.3%	44.5	1.6%	48.8%	3.4	60	200	232	25	14	59	23
Diseases of the circulatory system	52.8%	22.3%	68.7	9.5%	60.9%	4.6	49	7,564	63,668	5,992	2,519	269,394	4,709
Diseases of the respiratory system	24.1%	4.4%	62.8	16.7%	48.9%	4.8	53	5,258	20,204	1,865	801	2,628	801
Diseases of the digestive system	20.0%	4.0%	55.2	12.0%	51.2%	4.7	55	10,359	19,343	1,717	873	1,523	1,013
Diseases of the skin and subcutaneous tissue	10.0%	0.5%	59.5	7.2%	48.3%	4.8	51	1,371	1,009	103	60	234	82
Diseases of the musculoskeletal system and connective tissue	19.4%	4.1%	66.0	4.1%	34.8%	4.8	47	10,750	3,104	1,090	862	2,359	699
Diseases of the genitourinary system	24.4%	7.2%	63.4	10.5%	58.0%	5.2	82	5,000	12,307	2,546	691	8,921	171,360
Pregnancy, childbirth and the puerperium	0.5%	0.2%	31.0	0.0%	0.0%	3.8	44	159	182	8	60	28	18
Certain conditions originating in the perinatal period	0.1%	0.0%	0.2	0.0%	60.0%	6.0	239	1	874	1	4	13	0
Congenital malformations, deformations and chromosomal abnormalities	3.1%	0.7%	8.2	6.3%	54.3%	3.6	73	1,262	9,230	9	69	285	51
Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	49.2%	2.1%	55.4	14.1%	50.8%	4.6	60	9,824	9,086	482	460	1,034	387
Injury, poisoning and certain other consequences of external causes	22.3%	6.8%	71.5	9.7%	39.5%	4.7	50	2,986	15,169	1,813	1,789	5,668	1,055
Factors influencing health status and contact with health services	31.7%	3.6%	30.5	2.1%	55.3%	3.9	67	3,417	30,951	1,388	447	3,234	636

[a] Prevalence of ICD10-chapters among the total population. All other columns apply for beneficiaries with the selected most cost-incurring ICD10-chapter per row.

TABLE 4 Top-1% beneficiaries according to age group, and total expenditure per ICD10-chapter†.

Age group	<1	1-10	11-20	21-30	31-40	41-50	51-60	61-70	71-80	>80
Number	938	1125	1793	1892	2506	4670	6856	9811	9815	5801
Average per capita costs	€ 81,458	€ 69,576	€ 68,220	€ 62,428	€ 59,161	€ 59,546	€ 55,569	€ 55,253	€ 52,123	€ 47,166
	Factors influencing health status and contact with health services 13.5%	Neoplasms 15.5%	Mental and behavioral disorders 21.2%	Mental and behavioral disorders 18.8%	Mental and behavioral disorders 14.1%	Neoplasms 12.9%	Neoplasms 16.0%	Neoplasms 17.2%	Diseases of the circulatory system 20.1%	Diseases of the circulatory system 21.1%
	Congenital malformations, deformations and chromosomal abnormalities 5.2%	Mental and behavioral disorders 11.0%	Neoplasms 9.1%	Neoplasms 5.8%	Neoplasms 8.9%	Mental and behavioral disorders 9.1%	Diseases of the circulatory system 9.9%	Diseases of the circulatory system 14.7%	Neoplasms 11.9%	Injury, poisoning and certain other consequences of external causes 15.0%
	Diseases of the digestive system 2.5%	Factors influencing health status and contact with health services 4.6%	Factors influencing health status and contact with health services 2.1%	Diseases of the genitourinary system 4.0%	Diseases of the genitourinary system 6.1%	Diseases of the genitourinary system 7.3%	Diseases of the genitourinary system 8.1%	Diseases of the genitourinary system 7.8%	Diseases of the genitourinary system 5.5%	Diseases of the genitourinary system 9.8%
	Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified 1.8%	Congenital malformations, deformations and chromosomal abnormalities 2.9%	Injury, poisoning and certain other consequences of external causes 1.6%	Injury, poisoning and certain other consequences of external causes 2.2%	Diseases of the circulatory system 3.5%	Diseases of the circulatory system 5.9%	Mental and behavioral disorders 5.2%	Injury, poisoning and certain other consequences of external causes 2.5%	Injury, poisoning and certain other consequences of external causes 5.1%	Neoplasms 6.1%
	Neoplasms 1.6%	Diseases of the nervous system 2.4%	Diseases of the genitourinary system 1.5%	Factors influencing health status and contact with health services 2.2%	Factors influencing health status and contact with health services 2.6%	Factors influencing health status and contact with health services 2.6%	Factors influencing health status and contact with health services 2.6%	Diseases of the respiratory system 2.5%	Diseases of the musculoskeletal system and connective tissue 2.9%	Diseases of the musculoskeletal system and connective tissue 4.9%

† Total costs per ICD-chapter were summed per age group. In the table, the five ICD10-chapters with highest costs per age group are presented. I.e. among beneficiaries 1-10 years old, 15.5% of total costs were accounted for by neoplasm care.

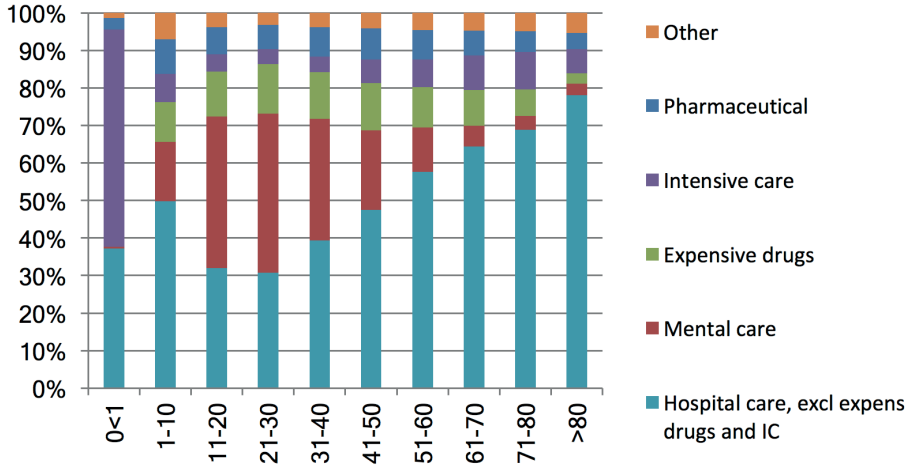


FIGURE 1 Cost drivers per age group in top 1% beneficiaries.

Discussion

In this study, we determined medical needs, demographics and utilization patterns of high-cost beneficiaries in the Netherlands. Expensive treatments, most cost-incurring condition, and age proved to be informative variables for studying this heterogeneous population. We found that expensive care use (expensive drugs, ICU treatment, dialysis, transplant care, DRG > €30,000) contributed to high costs in one third of top-1% beneficiaries and in less than 10% of top-2-5% beneficiaries. High-cost beneficiaries were overwhelmingly treated for diseases of circulatory system, neoplasms, and mental disorders. However, neoplasms and mental disorders were mainly found as most cost-incurring condition for a beneficiary, whereas circulatory disorders were mainly found as secondary condition. More than 50% of high-cost beneficiaries were 65 years of age or younger, and average costs decreased sharply with higher age within the top-1% population. Such insights are needed to develop tailored interventions and inform policy aimed at the high-need, high-cost populations.

Strengths and limitations

This was the first study assessing utilization patterns of high-cost beneficiaries in a European universal health system, and we used innovative variables to examine characteristics and utilization. We used data from one health insurer with a market share of approximately 27%, with data representative for the Dutch population. Despite the limited number of variables, our data allowed detailed identification of health care use and categorization of costs towards conditions. We chose to use expensive treatments, most cost-incurring condition and age as variables for further analyses as such analyses were lacking in the literature

and we regarded these most informative for policy and practice. One limitation is that our analysis was restricted to one year only. Consequently, we could not discern persistent high-cost users from episodic high-cost users (those with a single high-cost event [5]).

Reflections on our findings

Our findings generally align with prior research on high-cost beneficiaries. Similar to US studies [12,22], we identified three main subgroups of high-cost beneficiaries with cardiovascular diseases, mental disorders, and neoplasms, as well as several smaller subgroups. In addition, our findings confirm that high-cost beneficiaries are usually treated for several conditions and use care from multiple providers [10]. Like prior studies [12,22] we reported a high prevalence of diabetes, but this condition had a limited direct cost impact. This may be explained by the fact that Dutch diabetic care is primarily situated in primary care. Moreover, complications of diabetes were aggregated to the particular condition (e.g. retinopathy) using our link table. Furthermore, in line with Aldridge et al [5], we found that dying increases the risk for high costs (data not shown), but that less than ten percent of high-cost beneficiaries were in their last year of life. However, we also found that 64% of those dying did not incur high costs, compared to 80% of decedents in the US who did incur high costs [5]. This may be explained by decedents that could have used long term care services which were not included in our analyses. However, this may also result from the GP oriented organization of palliative care in the Netherlands, which is known for its low costs [23,24].

Our study is unique in estimating the relative contribution of expensive treatments in high-cost beneficiaries. The findings indicate that high unit costs for selected services play a substantial role in high-costs beneficiaries. We identified expensive treatment users among expensive patients. Furthermore, our analyses show expensive treatment users may use a lot of care besides such expensive treatments, suggesting that better alignment of expensive treatments with other care may be worthwhile. In line with Joynt et al, we suggest that expensive procedures (including orthopedic surgery, pacemaker-implantation etc) and catastrophic events may be a more significant cost driver in high-cost beneficiaries than avoidable hospitalizations, and that a complementary approach (see below) in high-need high-cost programs is needed [22].

To our knowledge, we are the first that have distinguished the most cost-incurring versus secondary conditions in high-cost beneficiaries. For example, diseases of circulatory system were mainly found as a secondary condition, though they also frequently occurred as most cost-incurring condition. In addition, mental disorders and neoplasms were predominantly the most cost-incurring condition. Our findings contribute to the rapidly evolving field of multimorbidity and patterns of healthcare use. We suggest that conditions that were frequently and primarily found as most cost-incurring condition should be priorities for policies that seek to contain costs and improve quality of care. However, the observational nature of our study does not allow for causal inference; i.e. the high number of morbidities in cancer patients may either indicate the many complications from cancer treatment, or point to prior chronic disease in patients with cancer.

Many high-cost beneficiaries were 65 years of age or younger; and the average costs decreased sharply with increasing age within the top-1% beneficiaries. In addition, we found typical care needs and utilization per age group. Both findings have rarely been

reported in literature [10] and underline the need for studies in the general population with comprehensive data. Furthermore, high-need, high-cost programs need to be aimed at beneficiaries of all ages; a mere focus on elderly would leave many high-cost beneficiaries unaddressed.

Policy and research implications

Our findings suggest a need for approaches that address patients' care needs across multiple conditions and to integrate care use across multiple providers. Important policy questions remain concerning the breadth of health care delivery innovations (i.e. care coordination programs, bundled payments; what should a bundle encompass?) [25]. We suggest that high-need, high-cost programs may aim to align the usual care for most cost-incurring conditions with the care for associated or common secondary conditions in specific care pathways. Furthermore, based on our findings we recommend a complementary approach geared towards expensive procedures and drugs as well as the extensive additional care besides expensive treatments. This suggests bundled payments may be worthwhile, as well as multidisciplinary assessment of patients' care needs for expensive treatments. In addition, prices for expensive drugs or procedures could be lowered, for example through reference pricing or competitive bidding [26,27].

Our research provides a precise picture of high-cost beneficiaries, but further research is necessary to specify characteristics and utilization of high-cost beneficiaries at a local level. Patient segmentation analysis has been suggested as a method for identifying homogenous target population groups from diverse populations, which allows for tailored policies [28]. Our analyses may inform such segmentation analyses. Furthermore, we suggest research into longitudinal patterns of multimorbidity to identify relevant subgroups that benefit from intervention. More research is needed to identify beneficiaries at risk of incurring high costs [29].

In conclusion, our findings show that high-cost beneficiaries are usually treated for several conditions and use care from multiple providers. Expensive treatments, the most cost-incurring condition, and age proved to be informative variables for studying this heterogeneous population. Tailored interventions are needed to meet the needs of high-cost beneficiaries, and to avoid waste of scarce resources.

References

1 Blumenthal, D. and M.K. Abrams, Tailoring Complex Care Management for High-Need, High-Cost Patients. JAMA, 2016.
2 Wennberg, J.E., et al., Inpatient care intensity and patients' ratings of their hospital experiences. Health Aff (Millwood), 2009. 28(1): p. 103-12.
3 Bodenheimer, T. and A. Fernandez, High and rising health care costs. Part 4: can costs be controlled while preserving quality? Ann Intern Med, 2005. 143(1): p. 26-31.
4 Colla, C.H., et al., Association Between Medicare Accountable Care Organization Implementation and Spending Among Clinically Vulnerable Beneficiaries. JAMA Intern Med, 2016. 176(8): p. 1167-75.
5 Aldridge, M.D. and A.S. Kelley, The Myth Regarding the High Cost of End-of-Life Care. American Journal of Public Health, 2015. 105(12): p. 2411-2415.
6 Hayes, S.L., et al., High-Need, High-Cost Patients: Who Are They and How Do They Use Health Care? A Population-Based Comparison of Demographics, Health Care Use, and Expenditures. Issue Brief (Commonw Fund), 2016. 26: p. 1-14.
7 Hwang, W., et al., Persistent high utilization in a privately insured population. Am J Manag Care, 2015. 21(4): p. 309-16.
8 Zulman, D.M., et al., Multimorbidity and healthcare utilisation among high-cost patients in the US Veterans Affairs Health Care System. BMJ Open, 2015. 5(4): p. e007771.
9 Lehnert, T., et al., Review: health care utilization and costs of elderly persons with multiple chronic conditions. Med Care Res Rev, 2011. 68(4): p. 387-420.
10 Wodchis, W.P., P.C. Austin, and D.A. Henry, A 3-year study of high-cost users of health care. CMAJ, 2016. 188(3): p. 182-8.
11 Monheit, A.C., Persistence in health expenditures in the short run: prevalence and consequences. Med Care, 2003. 41(7 Suppl): p. III53-III64.
12 Riley, G.F., Long-term trends in the concentration of Medicare spending. Health Aff (Millwood), 2007. 26(3): p. 808-16.
13 Rosella, L.C., et al., High-cost health care users in Ontario, Canada: demographic, socio-economic, and health status characteristics. BMC Health Serv Res, 2014. 14: p. 532.
14 Coughlin, T.A. and S.K. Long, Health care spending and service use among high-cost Medicaid beneficiaries, 2002-2004. Inquiry, 2009. 46(4): p. 405-17.
15 Kroneman, M., et al., The Netherlands: health system review. Vol. 18(2). 2016.
16 Mossialios, E., et al., 2015 International Profiles of Health Care Systems, C. Fund, Editor. 2016.
17 Smeets, H.M., N.J. de Wit, and A.W. Hoes, Routine health insurance data for scientific research: potential and limitations of the Agis Health Database. J Clin Epidemiol, 2011. 64(4): p. 424-30.
18 WHO. International Statistical Classification of Diseases and Related Health Problems 10th Revision. 2016 2016-10-23]; Available from: http://apps.who.int/classifications/icd10/browse/2016/en.

19 van Veen; S.H.C.M., Evaluating and Improving the Predictive Performance of Risk Equalization Models in Health Insurance Markets. 2016, Erasmus University Rotterdam: Rotterdam.
20 NZa, Investigation of the accessibility and affordability of medicines in specialist medical care. 2015, Dutch Healthcare Authority.
21 NZa. Open DIS data. 2016 2016-10-23]; Available from: www.opendisdata.nl
22 Joynt, K.E., et al., Contribution of preventable acute care spending to total spending for high-cost Medicare patients. JAMA, 2013. 309(24): p. 2572-8.
23 Evans, N., et al., End-of-life communication: a retrospective survey of representative general practitioner networks in four countries. J Pain Symptom Manage, 2014. 47(3): p. 604-619 e3.
24 Bekelman, J.E., et al., Comparison of Site of Death, Health Care Utilization, and Hospital Expenditures for Patients Dying With Cancer in 7 Developed Countries. JAMA, 2016. 315(3): p. 272-83.
25 Herzlinger, R.E., S.M. Schleicher, and S. Mullangi, Health Care Delivery Innovations That Integrate Care? Yes!: But Integrating What? JAMA, 2016. 315(11): p. 1109-10.
26 Acosta, A., et al., Pharmaceutical policies: effects of reference pricing, other pricing, and purchasing policies. Cochrane Database Syst Rev, 2014(10): p. CD005979.
27 Stadhouders, N., et al., Policy options to contain healthcare costs: a review and classification. Health Policy, 2016. 120(5): p. 486-94.
28 Vuik, S.I., E.K. Mayer, and A. Darzi, Patient Segmentation Analysis Offers Significant Benefits For Integrated Care And Support. Health Aff (Millwood), 2016. 35(5): p. 769-75.
29 Vrijhoef, B. and R. Thorlby, Developing care for a changing population: supporting patients with costly, complex needs. 2016, Nuffield Trust.

Appendix 1a. Five parameters for top-1% beneficiaries

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
A00-A09 Intestinal infectious diseases	0,9%	0,1%	8,9%	19,4%	0,1%
A15-A19 Tuberculosis	0,2%	0,0%	23,4%	36,6%	0,0%
A20-A28 Certain zoonotic bacterial diseases	0,0%				0,0%
A30-A49 Other bacterial diseases	2,9%	0,7%	22,7%	23,8%	0,3%
A50-A64 Infections with a predominantly sexual mode of transmission	0,1%				0,0%
A65-A69 Other spirochaetal diseases	0,0%				0,0%
B00-B09 Viral infections characterized by skin and mucous membrane lesions	0,4%				0,0%
B15-B19 Viral hepatitis	0,3%	0,1%	18,5%	10,4%	0,0%
B20-B24 Human immunodeficiency virus [HIV] disease	5,0%	0,2%	4,7%	16,8%	0,1%
B25-B34 Other viral diseases	0,3%	0,0%	8,6%	12,2%	0,0%
B50-B64 Protozoal diseases	0,0%				
B85-B89 Pediculosis, acariasis and other infestations	0,0%				0,0%
B99-B99 Other infectious diseases	0,9%	0,1%	12,5%	22,5%	0,1%
C00-C14 Malignant neoplasms of lip, oral cavity and pharynx	1,2%	0,5%	42,4%	40,8%	0,2%
C15-C26 Malignant neoplasms of digestive organs	7,5%	5,4%	71,2%	47,9%	2,4%
C30-C39 Malignant neoplasms of respiratory and intrathoracic organs	5,9%	3,5%	59,1%	52,5%	1,7%
C40-C41 Malignant neoplasms of bone and articular cartilage	0,4%	0,1%	28,1%	56,0%	0,2%
C43-C44 Melanoma and other malignant neoplasms of skin	3,0%	0,3%	9,9%	16,2%	0,1%
C45-C49 Malignant neoplasms of mesothelial and soft tissue	0,7%	0,3%	44,1%	36,8%	0,2%
C50-C50 Malignant neoplasm of breast	5,0%	3,7%	75,4%	42,8%	1,4%
C51-C58 Malignant neoplasms of female genital organs	1,3%	0,8%	62,0%	50,0%	0,4%
C60-C63 Malignant neoplasms of male genital organs	2,2%	0,8%	37,9%	34,8%	0,3%
C64-C68 Malignant neoplasms of urinary tract	3,5%	1,3%	36,9%	44,6%	0,6%
C69-C72 Malignant neoplasms of eye, brain and other parts of central nervous system	1,1%	0,2%	14,7%	41,1%	0,2%
C73-C75 Malignant neoplasms of thyroid and other endocrine glands	0,2%	0,1%	31,8%	50,1%	0,1%
C76-C80 Malignant neoplasms of ill-defined, secondary and unspecified sites	4,2%	1,1%	25,6%	40,4%	0,6%
C81-C96 Malignant neoplasms, stated or presumed to be primary, of lymphoid, haemato-poietic and related tissue	5,6%	4,1%	74,3%	41,4%	3,0%
D00-D09 In situ neoplasms	1,5%	0,0%	0,4%	2,6%	0,0%
D10-D36 Benign neoplasms	4,5%	0,2%	3,4%	20,8%	0,1%
D37-D48 Neoplasms of uncertain or unknown behaviour	3,2%	1,1%	32,5%	41,6%	0,7%

D50-D53 Nutritional anaemias	1,4%	0,1%	5,5%	22,7%	0,1%
D55-D59 Haemolytic anaemias	0,3%	0,1%	31,8%	25,2%	0,0%
D60-D64 Aplastic and other anaemias	0,5%	0,1%	12,7%	35,1%	0,1%
D65-D69 Coagulation defects, purpura and other haemorrhagic conditions	1,2%	0,5%	39,4%	6,1%	0,1%
D70-D77 Other diseases of blood and blood-forming organs	0,4%	0,0%	12,9%	28,5%	0,0%
D80-D89 Certain disorders involving the immune mechanism	0,7%	0,3%	41,4%	15,1%	0,1%
E00-E07 Disorders of thyroid gland	4,2%	0,0%	0,7%	16,0%	0,0%
E10-E14 Diabetes mellitus	13,8%	0,3%	2,1%	24,2%	0,2%
E20-E35 Disorders of other endocrine glands	1,1%	0,2%	19,5%	6,6%	0,0%
E40-E46 Malnutrition	1,5%	0,1%	7,6%	11,6%	0,0%
E50-E64 Other nutritional deficiencies	0,0%	0,0%	14,4%	12,3%	0,0%
E65-E68 Obesity and other hyperalimentation	0,4%	0,1%	24,4%	23,8%	0,0%
E70-E90 Metabolic disorders	2,6%	0,7%	25,9%	18,1%	0,2%
F00-F09 Organic, including symptomatic, mental disorders	6,5%	0,6%	9,7%	20,9%	0,2%
F10-F19 Mental and behavioural disorders due to psychoactive substance use	5,0%	2,7%	54,0%	34,0%	0,9%
F20-F29 Schizophrenia, schizotypal and delusional disorders	5,7%	3,5%	60,7%	32,9%	1,1%
F30-F39 Mood [affective] disorders	12,7%	3,0%	23,5%	35,4%	1,1%
F40-F48 Neurotic, stress-related and somatoform disorders	2,8%	1,1%	39,0%	43,2%	0,5%
F50-F59 Behavioural syndromes associated with physiological disturbances and physical factors	0,6%	0,3%	52,3%	51,9%	0,2%
F60-F69 Disorders of adult personality and behaviour	2,4%	1,4%	58,5%	42,1%	0,6%
F70-F79 Mental retardation	0,1%	0,0%	12,6%	8,4%	0,0%
F80-F89 Disorders of psychological development	1,3%	0,9%	68,7%	46,3%	0,4%
F90-F98 Behavioural and emotional disorders with onset usually occurring in childhood and adolescence	1,6%	0,7%	41,8%	58,0%	0,3%
F99-F99 Unspecified mental disorder	13,1%	0,6%	4,8%	32,7%	0,5%
G00-G09 Inflammatory diseases of the central nervous system	0,8%	0,2%	27,5%	30,7%	0,1%
G10-G14 Systemic atrophies primarily affecting the central nervous system	0,2%	0,0%	17,7%	16,1%	0,0%
G20-G26 Extrapyramidal and movement disorders	9,2%	0,2%	2,3%	12,0%	0,0%
G35-G37 Demyelinating diseases of the central nervous system	0,5%	0,2%	37,4%	14,0%	0,0%
G40-G47 Episodic and paroxysmal disorders	8,4%	0,7%	8,5%	33,8%	0,4%
G50-G59 Nerve, nerve root and plexus disorders	2,0%	0,0%	2,2%	22,0%	0,0%
G60-G64 Polyneuropathies and other disorders of the peripheral nervous system	1,5%	0,3%	19,4%	15,6%	0,1%
G70-G73 Diseases of myoneural junction and muscle	0,7%	0,2%	24,8%	32,2%	0,1%
G80-G83 Cerebral palsy and other paralytic syndromes	0,0%	0,0%	21,6%	3,9%	0,0%
G90-G99 Other disorders of the nervous system	3,2%	0,4%	12,2%	34,7%	0,2%
H00-H06 Disorders of eyelid, lacrimal system and orbit	1,1%	0,0%	1,2%	3,1%	0,0%

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
H10-H13 Disorders of conjunctiva	1,1%				0,0%
H15-H22 Disorders of sclera, cornea, iris and ciliary body	0,7%	0,0%	1,5%	6,5%	0,0%
H25-H28 Disorders of lens	4,1%	0,1%	2,7%	4,4%	0,1%
H30-H36 Disorders of choroid and retina	5,7%	0,1%	1,2%	24,5%	0,1%
H40-H42 Glaucoma	3,3%	0,0%	0,3%	1,4%	0,0%
H43-H45 Disorders of vitreous body and globe	0,6%	0,0%	0,4%	0,1%	0,0%
H46-H48 Disorders of optic nerve and visual pathways	0,9%	0,0%	0,2%	0,2%	0,0%
H49-H52 Disorders of ocular muscles, binocular movement, accommodation and refraction	0,9%	0,0%	1,3%	9,3%	0,0%
H53-H54 Visual disturbances and blindness	0,4%	0,0%	0,6%	0,6%	0,0%
H55-H59 Other disorders of eye and adnexa	2,2%	0,0%	0,4%	0,6%	0,0%
H60-H62 Diseases of external ear	1,5%	0,0%	1,5%	33,9%	0,0%
H65-H75 Diseases of middle ear and mastoid	1,4%	0,0%	3,5%	9,0%	0,0%
H80-H83 Diseases of inner ear	0,7%	0,0%	1,6%	7,6%	0,0%
H90-H95 Other disorders of ear	2,3%	0,2%	9,8%	81,4%	0,2%
I00-I02 Acute rheumatic fever	0,0%				0,0%
I05-I09 Chronic rheumatic heart diseases	0,4%	0,4%	95,0%	50,8%	0,2%
I10-I15 Hypertensive diseases	6,4%	0,0%	0,5%	15,0%	0,0%
I20-I25 Ischaemic heart diseases	12,7%	3,7%	29,0%	41,9%	1,6%
I26-I28 Pulmonary heart disease and diseases of pulmonary circulation	1,5%	0,3%	18,1%	13,7%	0,1%
I30-I33 Pericarditis/endocarditis	1,3%	0,2%	18,1%	42,4%	0,2%
I34-I39 Valve disorders	6,1%	0,7%	11,0%	62,5%	0,5%
I40-I41 Myocarditis	0,1%	0,0%	20,1%	19,3%	0,0%
I44-I49 Atrial fibrillation, rhythm and conduction disorders	11,8%	2,9%	24,3%	58,5%	1,6%
I50 Heart failure	9,3%	2,6%	28,4%	57,1%	1,5%
I51-I52 Complications/ill-defined descriptions, other heart disorders	9,6%	3,2%	33,1%	50,3%	1,6%
I60-I69 Cerebrovascular diseases	7,9%	4,2%	53,1%	52,7%	2,1%
I70-I79 Diseases of arteries, arterioles and capillaries	9,6%	4,1%	42,7%	47,3%	2,0%
I80-I89 Diseases of veins, lymphatic vessels and lymph nodes, not elsewhere classified	5,9%	0,1%	1,2%	15,4%	0,2%
I95-I99 Other and unspecified disorders of the circulatory system	5,7%	0,1%	1,2%	30,6%	0,1%
J00-J06 Acute upper respiratory infections	0,6%	0,0%	5,1%	18,2%	0,0%
J09-J18 Influenza and pneumonia	6,4%	1,2%	19,0%	20,4%	0,5%
J20-J22 Other acute lower respiratory infections	0,8%	0,1%	15,6%	20,0%	0,1%
J30-J39 Other diseases of upper respiratory tract	1,8%	0,1%	3,6%	13,1%	0,0%

J40-J47 Chronic lower respiratory diseases	14,1%	1,8%	13,1%	26,2%	0,6%
J60-J70 Lung diseases due to external agents	0,0%				
J80-J84 Other respiratory diseases principally affecting the interstitium	0,8%	0,2%	19,1%	24,5%	0,1%
J85-J86 Suppurative and necrotic conditions of lower respiratory tract	0,2%	0,0%	17,7%	25,1%	0,0%
J90-J94 Other diseases of pleura	2,2%	0,2%	8,4%	33,6%	0,1%
J95-J99 Other diseases of the respiratory system	2,9%	0,5%	15,8%	42,2%	0,3%
K00-K14 Diseases of oral cavity, salivary glands and jaws	0,4%	0,0%	1,8%	6,4%	0,0%
K20-K31 Diseases of oesophagus, stomach and duodenum	3,9%	0,3%	6,6%	18,7%	0,2%
K35-K38 Diseases of appendix	0,2%	0,0%	23,3%	21,9%	0,0%
K40-K46 Hernia	1,6%	0,1%	8,6%	17,1%	0,1%
K50-K52 Noninfective enteritis and colitis	2,4%	0,8%	32,4%	23,6%	0,2%
K55-K64 Other diseases of intestines	5,0%	0,9%	18,4%	31,3%	0,4%
K65-K67 Diseases of peritoneum	1,0%	0,3%	31,7%	27,1%	0,1%
K70-K77 Diseases of liver	1,7%	0,4%	24,8%	43,1%	0,3%
K80-K87 Disorders of gallbladder, biliary tract and pancreas	2,2%	0,5%	24,0%	34,8%	0,3%
K90-K93 Other diseases of the digestive system	7,0%	0,7%	9,8%	26,5%	0,4%
L00-L08 Infections of the skin and subcutaneous tissue	3,4%	0,3%	9,3%	25,1%	0,2%
L10-L14 Bullous disorders	0,7%	0,0%	1,7%	15,0%	0,0%
L20-L30 Dermatitis and eczema	2,4%	0,0%	0,7%	20,7%	0,0%
L40-L45 Papulosquamous disorders	0,7%	0,1%	10,2%	10,2%	0,0%
L50-L54 Urticaria and erythema	0,1%	0,0%	4,7%	1,2%	0,0%
L60-L75 Disorders of skin appendages	0,6%	0,0%	1,1%	12,0%	0,0%
L80-L99 Other disorders of the skin and subcutaneous tissue	3,2%	0,1%	4,6%	27,8%	0,1%
M00-M03 Infectious arthropathies	0,1%	0,0%	23,2%	31,7%	0,0%
M05-M14 Inflammatory polyarthropathies	5,0%	0,4%	8,0%	22,6%	0,1%
M15-M19 Arthrosis	4,5%	1,7%	37,4%	51,4%	0,8%
M20-M25 Other joint disorders	2,3%	0,3%	12,2%	34,4%	0,1%
M30-M36 Systemic connective tissue disorders	1,5%	0,3%	18,0%	26,9%	0,1%
M40-M43 Deforming dorsopathies	0,4%	0,1%	34,1%	55,1%	0,1%
M45-M49 Spondylopathies	1,7%	0,3%	17,8%	28,9%	0,1%
M50-M54 Other dorsopathies	3,9%	0,5%	12,2%	39,2%	0,2%
M60-M63 Disorders of muscles	0,1%	0,0%	1,6%	2,3%	0,0%
M65-M68 Disorders of synovium and tendon	0,3%	0,0%	2,0%	12,3%	0,0%
M70-M79 Other soft tissue disorders	1,9%	0,1%	3,3%	12,7%	0,0%
M80-M85 Disorders of bone density and structure	1,4%	0,0%	1,3%	20,8%	0,0%
M86-M90 Other osteopathies	1,0%	0,1%	12,2%	33,1%	0,1%
M91-M94 Chondropathies	0,0%	0,0%	12,6%	82,9%	0,0%

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
M95-M99 Other disorders of the musculoskeletal system and connective tissue	0,1%	0,0%	5,5%	56,1%	0,0%
N00-N08 Glomerular diseases	0,3%	0,0%	14,2%	31,0%	0,0%
N10-N16 Renal tubulo-interstitial diseases	1,1%	0,1%	11,6%	30,1%	0,1%
N17-N19 Renal failure	12,2%	6,4%	52,4%	66,0%	6,8%
N20-N23 Urolithiasis	0,8%	0,1%	14,4%	28,3%	0,1%
N25-N29 Other disorders of kidney and ureter	1,8%	0,1%	4,1%	21,0%	0,1%
N30-N39 Other diseases of urinary system	6,5%	0,4%	5,7%	21,9%	0,2%
N40-N51 Diseases of male genital organs	3,4%	0,1%	1,9%	15,9%	0,1%
N60-N64 Disorders of breast	0,6%	0,0%	1,1%	7,7%	0,0%
N70-N77 Inflammatory diseases of female pelvic organs	0,4%	0,0%	3,0%	3,9%	0,0%
N80-N98 Noninflammatory disorders of female genital tract	2,4%	0,1%	3,0%	12,0%	0,0%
O00-O08 Pregnancy with abortive outcome	0,1%	0,0%	5,2%	1,1%	0,0%
O20-O29 Other maternal disorders predominantly related to pregnancy	0,0%				0,0%
O60-O75 Complications of labour and delivery	0,0%				0,0%
O80-O84 Delivery	0,4%	0,0%	7,6%	27,0%	0,0%
O94-O99 Other obstetric conditions, not elsewhere classified	0,4%	0,2%	43,9%	59,1%	0,1%
P20-P29 Respiratory and cardiovascular disorders specific to the perinatal period	0,0%	0,0%	23,9%	9,2%	0,0%
P50-P61 Haemorrhagic and haematological disorders of fetus and newborn	0,0%	0,0%	25,1%	26,9%	0,0%
Q00-Q07 Congenital malformations of the nervous system	0,2%	0,0%	17,7%	16,8%	0,0%
Q10-Q18 Congenital malformations of eye, ear, face and neck	1,0%	0,0%	0,7%	1,2%	0,0%
Q20-Q28 Congenital malformations of the circulatory system	1,2%	0,5%	42,4%	39,8%	0,2%
Q30-Q34 Congenital malformations of the respiratory system	0,0%	0,0%	23,9%	27,9%	0,0%
Q35-Q37 Cleft lip and cleft palate	0,0%	0,0%	18,3%	24,6%	0,0%
Q38-Q45 Other congenital malformations of the digestive system	0,1%	0,0%	5,2%	53,9%	0,0%
Q50-Q56 Congenital malformations of genital organs	0,1%	0,0%	7,2%	6,6%	0,0%
Q60-Q64 Congenital malformations of the urinary system	0,1%	0,0%	6,6%	14,6%	0,0%
Q65-Q79 Congenital malformations and deformations of the musculoskeletal system	0,4%	0,0%	10,8%	34,6%	0,0%
Q80-Q89 Other congenital malformations	0,2%	0,0%	10,9%	13,4%	0,0%
Q90-Q99 Chromosomal abnormalities, not elsewhere classified	0,2%	0,0%	10,9%	9,1%	0,0%
R01 Cardiac murmurs and other cardiac sounds	0,0%	0,0%	15,1%	8,0%	0,0%
R04 Haemorrhage from respiratory passages	1,2%	0,0%	2,3%	19,3%	0,0%
R05 Cough	0,3%	0,0%	4,2%	11,1%	0,0%
R06 Abnormalities of breathing	3,8%	0,6%	16,4%	32,7%	0,3%
R07 Pain in throat and chest	0,5%	0,0%	1,8%	21,9%	0,0%

R09 Other symptoms and signs involving the circulatory and respiratory systems	0,0%	0,0%	5,0%	9,8%	0,0%
R10 Abdominal and pelvic pain	2,3%	0,1%	2,4%	21,5%	0,1%
R11 Nausea and vomiting	0,0%	0,0%	9,1%	30,3%	0,0%
R13 Dysphagia	1,3%	0,0%	0,9%	2,8%	0,0%
R15 Faecal incontinence	0,1%	0,0%	43,1%	43,1%	0,0%
R22.1 Localized swelling, mass and lump, neck	0,2%	0,0%	2,1%	11,6%	0,0%
R26 Abnormalities of gait and mobility	0,1%	0,0%	2,3%	40,9%	0,0%
R29 Other symptoms and signs involving the nervous and musculoskeletal systems	1,2%	0,0%	2,9%	14,3%	0,0%
R31 Unspecified haematuria	0,1%	0,0%	7,7%	5,7%	0,0%
R32 Unspecified urinary incontinence	0,0%	0,0%	7,7%	5,3%	0,0%
R35 Polyuria	0,0%	0,0%	50,3%	73,5%	0,0%
R39 Other symptoms and signs involving the urinary system	0,2%				0,0%
R40 Somnolence, stupor and coma	0,0%				0,0%
R42 Dizziness and giddiness	0,1%	0,0%	3,1%	44,1%	0,0%
R43 Disturbances of smell and taste	0,0%				0,0%
R47 Speech disturbances, not elsewhere classified	0,0%				0,0%
R49 Voice disturbances	1,2%	0,0%	0,9%	3,2%	0,0%
R50 Fever of other and unknown origin	1,1%	0,0%	3,6%	27,4%	0,1%
R51 Headache	0,0%	0,0%	5,3%	0,5%	0,0%
R52 Pain, not elsewhere classified	2,6%	0,1%	4,9%	45,9%	0,1%
R53 Malaise and fatigue	1,2%	0,1%	4,5%	16,2%	0,0%
R54 Senility	3,1%	0,7%	24,5%	31,8%	0,3%
R55 Syncope and collapse	1,5%	0,0%	3,2%	14,3%	0,0%
R56 Convulsions, not elsewhere classified	0,6%	0,1%	11,5%	40,4%	0,0%
R59 Enlarged lymph nodes	0,5%	0,0%	2,3%	52,9%	0,0%
R60 Oedema, not elsewhere classified	0,2%	0,0%	4,5%	10,2%	0,0%
R62 Lack of expected normal physiological development	0,1%	0,0%	23,2%	68,9%	0,0%
R63 Symptoms and signs concerning food and fluid intake	0,9%	0,1%	10,0%	28,0%	0,1%
R68 Other general symptoms and signs	12,8%	0,1%	0,5%	21,4%	0,1%
R69 Unknown and unspecified causes of morbidity	32,5%	0,3%	0,8%	13,7%	0,3%
R70-R79 Abnormal findings on examination of blood, without diagnosis	2,3%	0,1%	3,4%	22,1%	0,1%
R87 Abnormal findings in specimens from female genital organs	0,3%	0,0%	2,5%	5,2%	0,0%
R95-R99 Ill-defined and unknown causes of mortality	0,0%				0,0%
S00-S09 Injuries to the head	3,2%	0,4%	11,4%	29,4%	0,2%
S10-S19 Injuries to the neck	0,4%	0,0%	8,1%	21,9%	0,0%
S20-S29 Injuries to the thorax	0,6%	0,2%	27,9%	31,5%	0,1%
S30-S39 Injuries to the abdomen, lower back, lumbar spine and pelvis	0,8%	0,3%	41,6%	36,7%	0,1%

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
S40-S49 Injuries to the shoulder and upper arm	1,1%	0,1%	12,2%	19,3%	0,1%
S50-S59 Injuries to the elbow and forearm	0,3%	0,0%	7,3%	16,6%	0,0%
S60-S69 Injuries to the wrist and hand	1,2%	0,0%	4,0%	13,4%	0,0%
S70-S79 Injuries to the hip and thigh	4,3%	2,8%	64,2%	55,2%	1,4%
S80-S89 Injuries to the knee and lower leg	0,7%	0,2%	27,7%	17,3%	0,0%
S90-S99 Injuries to the ankle and foot	1,0%	0,2%	16,5%	16,6%	0,0%
T00-T07 Injuries involving multiple body regions	2,0%	0,5%	25,6%	39,1%	0,3%
T08-T14 Injuries to unspecified part of trunk, limb or body region	7,5%	0,3%	4,0%	26,1%	0,2%
T15-T19 Effects of foreign body entering through natural orifice	0,2%	0,0%	1,0%	0,3%	0,0%
T20-T32 Burns and corrosions	0,3%	0,1%	41,7%	61,2%	0,1%
T33-T35 Frostbite	0,0%				0,0%
T36-T50 Poisoning by drugs, medicaments and biological substances	1,5%	0,0%	1,3%	13,8%	0,0%
T51-T65 Toxic effects of substances chiefly nonmedicinal as to source	0,1%				0,0%
T66-T78 Other and unspecified effects of external causes	0,7%	0,1%	7,9%	9,4%	0,0%
T79-T79 Certain early complications of trauma	0,0%				0,0%
T80-T88 Complications of surgical and medical care, not elsewhere classified	2,0%	0,6%	32,1%	47,5%	0,4%
T90-T98 Sequelae of injuries, of poisoning and of other consequences of external causes	0,9%	0,1%	12,0%	24,6%	0,0%
Z00-Z13 Persons encountering health services for examination and investigation	5,5%	0,2%	4,2%	30,2%	0,2%
Z20-Z29 Persons with potential health hazards related to communicable diseases	0,0%				0,0%
Z30-Z39 Persons encountering health services in circumstances related to reproduction	3,9%	1,2%	29,5%	26,6%	0,4%
Z40-Z54 Persons encountering health services for specific procedures and health care	16,3%	2,0%	12,0%	42,4%	1,4%
Z80-Z99 Persons with potential health hazards related to family and personal history and certain conditions influencing health status	13,9%	0,3%	2,4%	56,2%	0,5%

Appendix 1b. Five parameters for top-2-5% beneficiaries

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
A00-A09 Intestinal infectious diseases	0,5%	0,2%	32,6%	32,5%	0,1%
A15-A19 Tuberculosis	0,1%	0,0%	22,5%	32,3%	0,0%
A20-A28 Certain zoonotic bacterial diseases	0,0%				
A30-A49 Other bacterial diseases	1,0%	0,6%	61,2%	39,8%	0,3%
A50-A64 Infections with a predominantly sexual mode of transmission	0,2%	0,0%	8,9%	9,2%	0,0%
A65-A69 Other spirochaetal diseases	0,0%	0,0%	14,8%	23,3%	0,0%
B00-B09 Viral infections characterized by skin and mucous membrane lesions	0,4%	0,0%	2,4%	4,8%	0,0%
B15-B19 Viral hepatitis	0,3%	0,1%	22,3%	13,8%	0,0%
B20-B24 Human immunodeficiency virus [HIV] disease	3,5%	2,2%	61,4%	13,3%	0,3%
B25-B34 Other viral diseases	0,1%	0,0%	29,4%	38,2%	0,0%
B50-B64 Protozoal diseases	0,0%	0,0%	50,6%	16,2%	0,0%
B85-B89 Pediculosis, acariasis and other infestations	0,0%	0,0%	23,8%	40,0%	0,0%
B99-B99 Other infectious diseases	0,3%	0,1%	42,5%	43,8%	0,1%
C00-C14 Malignant neoplasms of lip, oral cavity and pharynx	0,5%	0,2%	47,5%	45,8%	0,1%
C15-C26 Malignant neoplasms of digestive organs	3,8%	2,5%	67,2%	56,7%	1,8%
C30-C39 Malignant neoplasms of respiratory and intrathoracic organs	2,7%	1,6%	59,1%	52,1%	1,1%
C40-C41 Malignant neoplasms of bone and articular cartilage	0,1%	0,0%	28,6%	56,8%	0,0%
C43-C44 Melanoma and other malignant neoplasms of skin	3,0%	0,4%	12,2%	25,5%	0,2%
C45-C49 Malignant neoplasms of mesothelial and soft tissue	0,3%	0,2%	54,7%	47,6%	0,1%
C50-C50 Malignant neoplasm of breast	3,8%	2,5%	65,6%	60,1%	1,8%
C51-C58 Malignant neoplasms of female genital organs	0,9%	0,6%	64,3%	59,1%	0,4%
C60-C63 Malignant neoplasms of male genital organs	2,2%	0,7%	32,3%	53,2%	0,5%
C64-C68 Malignant neoplasms of urinary tract	2,8%	1,5%	53,6%	57,0%	1,0%
C69-C72 Malignant neoplasms of eye, brain and other parts of central nervous system	0,3%	0,1%	30,6%	50,3%	0,1%
C73-C75 Malignant neoplasms of thyroid and other endocrine glands	0,2%	0,1%	49,4%	57,0%	0,1%
C76-C80 Malignant neoplasms of ill-defined, secondary and unspecified sites	1,5%	0,5%	34,4%	46,6%	0,4%
C81-C96 Malignant neoplasms, stated or presumed to be primary, of lymphoid, haemato-poietic and related tissue	1,6%	0,8%	53,3%	40,1%	0,4%
D00-D09 In situ neoplasms	1,8%	0,1%	4,1%	10,8%	0,1%
D10-D36 Benign neoplasms	4,6%	0,5%	11,8%	34,2%	0,4%
D37-D48 Neoplasms of uncertain or unknown behaviour	1,6%	0,6%	38,4%	50,3%	0,4%
D50-D53 Nutritional anaemias	1,0%	0,2%	18,1%	33,2%	0,1%

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
D55-D59 Haemolytic anaemias	0,2%	0,1%	35,0%	37,6%	0,0%
D60-D64 Aplastic and other anaemias	0,3%	0,1%	21,0%	33,6%	0,0%
D65-D69 Coagulation defects, purpura and other haemorrhagic conditions	0,4%	0,1%	22,5%	32,3%	0,0%
D70-D77 Other diseases of blood and blood-forming organs	0,2%	0,0%	20,5%	34,0%	0,0%
D80-D89 Certain disorders involving the immune mechanism	0,3%	0,1%	36,0%	24,3%	0,0%
E00-E07 Disorders of thyroid gland	4,8%	0,1%	2,4%	34,3%	0,1%
E10-E14 Diabetes mellitus	12,2%	1,1%	9,3%	33,5%	0,5%
E20-E35 Disorders of other endocrine glands	1,1%	0,3%	31,5%	11,7%	0,1%
E40-E46 Malnutrition	0,3%	0,1%	16,8%	21,4%	0,0%
E50-E64 Other nutritional deficiencies	0,0%	0,0%	16,9%	40,7%	0,0%
E65-E68 Obesity and other hyperalimentation	1,4%	1,0%	70,5%	72,4%	0,6%
E70-E90 Metabolic disorders	2,3%	0,4%	16,1%	33,2%	0,2%
F00-F09 Organic, including symptomatic, mental disorders	3,0%	0,5%	17,3%	38,6%	0,3%
F10-F19 Mental and behavioural disorders due to psychoactive substance use	3,3%	1,8%	55,4%	51,5%	1,1%
F20-F29 Schizophrenia, schizotypal and delusional disorders	3,7%	2,1%	55,2%	59,0%	1,3%
F30-F39 Mood [affective] disorders	12,5%	2,7%	22,0%	49,8%	1,5%
F40-F48 Neurotic, stress-related and somatoform disorders	2,9%	1,6%	57,7%	53,9%	1,0%
F50-F59 Behavioural syndromes associated with physiological disturbances and physical factors	0,5%	0,3%	54,6%	65,7%	0,2%
F60-F69 Disorders of adult personality and behaviour	2,3%	1,6%	71,8%	57,7%	1,0%
F70-F79 Mental retardation	0,1%	0,0%	14,8%	11,6%	0,0%
F80-F89 Disorders of psychological development	1,0%	0,7%	70,8%	67,0%	0,5%
F90-F98 Behavioural and emotional disorders with onset usually occurring in childhood and adolescence	1,6%	0,8%	47,7%	74,0%	0,6%
F99-F99 Unspecified mental disorder	7,9%	1,0%	12,8%	43,6%	0,8%
G00-G09 Inflammatory diseases of the central nervous system	0,3%	0,1%	42,7%	50,3%	0,1%
G10-G14 Systemic atrophies primarily affecting the central nervous system	0,1%	0,0%	22,6%	21,3%	0,0%
G20-G26 Extrapyramidal and movement disorders	8,4%	0,2%	2,3%	27,7%	0,1%
G35-G37 Demyelinating diseases of the central nervous system	1,1%	0,6%	58,9%	10,7%	0,1%
G40-G47 Episodic and paroxysmal disorders	7,6%	1,3%	16,5%	33,7%	0,8%
G50-G59 Nerve, nerve root and plexus disorders	2,0%	0,2%	8,9%	23,4%	0,1%
G60-G64 Polyneuropathies and other disorders of the peripheral nervous system	0,8%	0,1%	10,3%	31,6%	0,1%
G70-G73 Diseases of myoneural junction and muscle	0,3%	0,1%	19,4%	30,3%	0,0%
G80-G83 Cerebral palsy and other paralytic syndromes	0,0%	0,0%	18,4%	7,5%	0,0%

G90-G99 Other disorders of the nervous system	1,6%	0,3%	16,2%	41,5%	0,2%
H00-H06 Disorders of eyelid, lacrimal system and orbit	1,4%	0,1%	4,2%	14,5%	0,0%
H10-H13 Disorders of conjunctiva	1,2%	0,0%	1,1%	6,2%	0,0%
H15-H22 Disorders of sclera, cornea, iris and ciliary body	0,9%	0,1%	8,8%	18,1%	0,0%
H25-H28 Disorders of lens	4,6%	0,5%	11,0%	16,6%	0,3%
H30-H36 Disorders of choroid and retina	6,3%	0,8%	12,3%	39,9%	0,4%
H40-H42 Glaucoma	3,8%	0,0%	1,3%	5,2%	0,0%
H43-H45 Disorders of vitreous body and globe	0,7%	0,0%	3,8%	19,2%	0,0%
H46-H48 Disorders of optic nerve and visual pathways	0,6%	0,0%	2,0%	10,4%	0,0%
H49-H52 Disorders of ocular muscles, binocular movement, accommodation and refraction	0,9%	0,0%	1,5%	11,1%	0,0%
H53-H54 Visual disturbances and blindness	0,4%	0,0%	1,2%	1,0%	0,0%
H55-H59 Other disorders of eye and adnexa	1,8%	0,0%	0,9%	1,2%	0,0%
H60-H62 Diseases of external ear	1,4%	0,1%	6,2%	37,9%	0,1%
H65-H75 Diseases of middle ear and mastoid	1,3%	0,2%	13,6%	43,5%	0,1%
H80-H83 Diseases of inner ear	0,7%	0,1%	7,1%	25,8%	0,0%
H90-H95 Other disorders of ear	1,9%	0,1%	3,5%	34,5%	0,1%
I00-I02 Acute rheumatic fever	0,0%	0,0%	28,9%	61,4%	0,0%
I05-I09 Chronic rheumatic heart diseases	0,0%	0,0%	72,3%	82,8%	0,0%
I10-I15 Hypertensive diseases	6,6%	0,1%	1,9%	23,2%	0,1%
I20-I25 Ischaemic heart diseases	11,0%	5,4%	49,1%	51,9%	3,3%
I26-I28 Pulmonary heart disease and diseases of pulmonary circulation	0,8%	0,3%	33,4%	34,0%	0,1%
I30-I33 Pericarditis/endocarditis	0,4%	0,1%	30,2%	50,9%	0,1%
I34-I39 Valve disorders	2,6%	0,2%	7,9%	35,8%	0,2%
I40-I41 Myocarditis	0,0%	0,0%	57,9%	36,0%	0,0%
I44-I49 Atrial fibrillation, rhythm and conduction disorders	8,9%	3,1%	35,0%	59,1%	2,2%
I50 Heart failure	4,6%	1,7%	36,7%	43,7%	1,0%
I51-I52 Complications/ill-defined descriptions, other heart disorders	3,3%	0,2%	7,2%	60,0%	0,3%
I60-I69 Cerebrovascular diseases	3,6%	2,2%	60,2%	55,4%	1,5%
I70-I79 Diseases of arteries, arterioles and capillaries	5,1%	2,0%	39,6%	59,0%	1,5%
I80-I89 Diseases of veins, lymphatic vessels and lymph nodes, not elsewhere classified	3,3%	0,3%	9,6%	26,9%	0,2%
I95-I99 Other and unspecified disorders of the circulatory system	3,7%	0,1%	3,7%	20,9%	0,2%
J00-J06 Acute upper respiratory infections	0,5%	0,0%	10,1%	21,0%	0,0%
J09-J18 Influenza and pneumonia	3,1%	1,6%	53,0%	38,4%	0,8%
J20-J22 Other acute lower respiratory infections	0,5%	0,2%	32,2%	32,4%	0,1%
J30-J39 Other diseases of upper respiratory tract	1,8%	0,2%	12,6%	32,6%	0,1%
J40-J47 Chronic lower respiratory diseases	13,6%	1,8%	13,6%	35,4%	0,9%

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
J60-J70 Lung diseases due to external agents	0,0%				
J80-J84 Other respiratory diseases principally affecting the interstitium	0,5%	0,2%	31,8%	40,1%	0,1%
J85-J86 Suppurative and necrotic conditions of lower respiratory tract	0,1%	0,0%	24,5%	47,7%	0,0%
J90-J94 Other diseases of pleura	0,9%	0,3%	36,7%	51,2%	0,2%
J95-J99 Other diseases of the respiratory system	1,3%	0,2%	17,1%	38,4%	0,2%
K00-K14 Diseases of oral cavity, salivary glands and jaws	0,2%	0,0%	2,7%	21,6%	0,0%
K20-K31 Diseases of oesophagus, stomach and duodenum	2,9%	0,5%	16,4%	32,0%	0,3%
K35-K38 Diseases of appendix	0,3%	0,2%	54,8%	47,3%	0,1%
K40-K46 Hernia	1,3%	0,4%	32,5%	39,5%	0,2%
K50-K52 Noninfective enteritis and colitis	2,6%	1,3%	50,0%	21,5%	0,4%
K55-K64 Other diseases of intestines	4,0%	1,1%	27,6%	42,5%	0,7%
K65-K67 Diseases of peritoneum	0,3%	0,2%	59,0%	49,0%	0,1%
K70-K77 Diseases of liver	1,0%	0,4%	35,7%	42,1%	0,2%
K80-K87 Disorders of gallbladder, biliary tract and pancreas	2,3%	1,4%	62,2%	53,2%	0,8%
K90-K93 Other diseases of the digestive system	3,5%	0,8%	22,0%	42,6%	0,5%
L00-L08 Infections of the skin and subcutaneous tissue	2,1%	0,5%	24,1%	38,1%	0,3%
L10-L14 Bullous disorders	0,5%	0,0%	4,6%	22,2%	0,0%
L20-L30 Dermatitis and eczema	2,0%	0,1%	3,9%	24,2%	0,1%
L40-L45 Papulosquamous disorders	1,1%	0,3%	28,4%	7,8%	0,0%
L50-L54 Urticaria and erythema	0,1%	0,0%	3,2%	32,7%	0,0%
L60-L75 Disorders of skin appendages	0,7%	0,0%	3,0%	9,3%	0,0%
L80-L99 Other disorders of the skin and subcutaneous tissue	2,4%	0,2%	8,4%	22,5%	0,1%
M00-M03 Infectious arthropathies	0,1%	0,0%	38,5%	44,5%	0,0%
M05-M14 Inflammatory polyarthropathies	6,6%	2,0%	31,1%	11,7%	0,4%
M15-M19 Arthrosis	9,3%	6,7%	72,4%	71,5%	4,5%
M20-M25 Other joint disorders	3,6%	0,7%	20,6%	41,2%	0,4%
M30-M36 Systemic connective tissue disorders	1,3%	0,3%	19,9%	34,0%	0,1%
M40-M43 Deforming dorsopathies	0,3%	0,1%	31,2%	68,8%	0,1%
M45-M49 Spondylopathies	2,4%	0,9%	37,5%	36,5%	0,4%
M50-M54 Other dorsopathies	5,5%	1,7%	30,3%	45,3%	1,0%
M60-M63 Disorders of muscles	0,2%	0,0%	6,0%	9,5%	0,0%
M65-M68 Disorders of synovium and tendon	0,5%	0,0%	6,5%	16,2%	0,0%
M70-M79 Other soft tissue disorders	2,6%	0,4%	14,5%	34,3%	0,2%
M80-M85 Disorders of bone density and structure	1,4%	0,1%	6,2%	18,9%	0,1%

M86-M90 Other osteopathies	1,0%	0,2%	19,4%	45,3%	0,1%
M91-M94 Chondropathies	0,1%	0,0%	27,4%	57,0%	0,0%
M95-M99 Other disorders of the musculoskeletal system and connective tissue	0,2%	0,0%	22,7%	45,3%	0,0%
N00-N08 Glomerular diseases	0,2%	0,1%	31,9%	45,5%	0,0%
N10-N16 Renal tubulo-interstitial diseases	0,7%	0,3%	38,2%	47,5%	0,2%
N17-N19 Renal failure	3,8%	1,1%	28,8%	36,4%	0,6%
N20-N23 Urolithiasis	1,0%	0,5%	45,7%	51,8%	0,3%
N25-N29 Other disorders of kidney and ureter	0,8%	0,1%	14,4%	39,5%	0,1%
N30-N39 Other diseases of urinary system	4,9%	1,0%	20,1%	28,5%	0,5%
N40-N51 Diseases of male genital organs	2,9%	0,5%	16,1%	38,0%	0,3%
N60-N64 Disorders of breast	0,9%	0,0%	4,8%	17,0%	0,0%
N70-N77 Inflammatory diseases of female pelvic organs	0,6%	0,0%	6,7%	27,8%	0,0%
N80-N98 Noninflammatory disorders of female genital tract	3,9%	1,0%	25,2%	42,8%	0,5%
O00-O08 Pregnancy with abortive outcome	0,5%	0,1%	21,0%	35,7%	0,1%
O20-O29 Other maternal disorders predominantly related to pregnancy	0,3%	0,0%	0,2%	15,5%	0,0%
O60-O75 Complications of labour and delivery	0,2%	0,0%	23,1%	28,4%	0,0%
O80-O84 Delivery	3,3%	1,6%	48,0%	35,2%	0,7%
O94-O99 Other obstetric conditions, not elsewhere classified	3,2%	1,3%	40,8%	45,9%	0,7%
P20-P29 Respiratory and cardiovascular disorders specific to the perinatal period	0,0%	0,0%	22,5%	24,5%	0,0%
P50-P61 Haemorrhagic and haematological disorders of fetus and newborn	0,0%	0,0%	35,4%	33,5%	0,0%
Q00-Q07 Congenital malformations of the nervous system	0,1%	0,0%	23,6%	24,0%	0,0%
Q10-Q18 Congenital malformations of eye, ear, face and neck	0,6%	0,0%	4,5%	21,5%	0,0%
Q20-Q28 Congenital malformations of the circulatory system	0,4%	0,2%	44,2%	46,4%	0,1%
Q30-Q34 Congenital malformations of the respiratory system	0,0%	0,0%	16,5%	50,5%	0,0%
Q35-Q37 Cleft lip and cleft palate	0,0%	0,0%	58,5%	41,8%	0,0%
Q38-Q45 Other congenital malformations of the digestive system	0,0%	0,0%	38,0%	54,5%	0,0%
Q50-Q56 Congenital malformations of genital organs	0,1%	0,0%	31,8%	50,0%	0,0%
Q60-Q64 Congenital malformations of the urinary system	0,1%	0,0%	25,4%	50,8%	0,0%
Q65-Q79 Congenital malformations and deformations of the musculoskeletal system	0,3%	0,1%	39,6%	56,5%	0,1%
Q80-Q89 Other congenital malformations	0,1%	0,0%	15,5%	15,2%	0,0%
Q90-Q99 Chromosomal abnormalities, not elsewhere classified	0,1%	0,0%	24,3%	9,4%	0,0%
R01 Cardiac murmurs and other cardiac sounds	0,0%	0,0%	23,0%	27,1%	0,0%
R04 Haemorrhage from respiratory passages	0,7%	0,1%	9,9%	27,6%	0,0%
R05 Cough	0,2%	0,0%	9,2%	16,1%	0,0%
R06 Abnormalities of breathing	1,8%	0,3%	17,2%	45,1%	0,2%
R07 Pain in throat and chest	0,3%	0,0%	6,7%	26,6%	0,0%
R09 Other symptoms and signs involving the circulatory and respiratory systems	0,0%	0,0%	41,6%	34,8%	0,0%

ICD10-subchapter (condition)	Prevalence	% as most cost incurring condition	% most cost incurring / prevalence	% of costs by most cost incurring condition	% of total costs
R10 Abdominal and pelvic pain	1,7%	0,2%	9,6%	30,7%	0,1%
R11 Nausea and vomiting	0,0%				0,0%
R13 Dysphagia	0,8%	0,0%	4,1%	17,7%	0,0%
R15 Faecal incontinence	0,1%	0,1%	45,9%	61,1%	0,0%
R22.1 Localized swelling, mass and lump, neck	0,1%	0,0%	7,8%	16,0%	0,0%
R26 Abnormalities of gait and mobility	0,1%	0,0%	13,7%	24,2%	0,0%
R29 Other symptoms and signs involving the nervous and musculoskeletal systems	1,1%	0,1%	6,1%	35,1%	0,1%
R31 Unspecified haematuria	0,0%	0,0%	8,3%	21,1%	0,0%
R32 Unspecified urinary incontinence	0,0%	0,0%	5,6%	2,3%	0,0%
R35 Polyuria	0,0%	0,0%	28,9%	19,2%	0,0%
R39 Other symptoms and signs involving the urinary system	0,2%	0,0%	9,4%	26,9%	0,0%
R40 Somnolence, stupor and coma	0,0%	0,0%	25,3%	25,4%	0,0%
R42 Dizziness and giddiness	0,1%	0,0%	9,8%	34,2%	0,0%
R43 Disturbances of smell and taste	0,0%	0,0%	4,0%	70,7%	0,0%
R47 Speech disturbances, not elsewhere classified	0,0%	0,0%	2,5%	11,9%	0,0%
R49 Voice disturbances	0,8%	0,0%	4,8%	20,0%	0,0%
R50 Fever of other and unknown origin	0,4%	0,1%	24,3%	38,8%	0,1%
R51 Headache	0,0%	0,0%	5,8%	36,0%	0,0%
R52 Pain, not elsewhere classified	1,2%	0,1%	10,4%	31,1%	0,1%
R53 Malaise and fatigue	0,8%	0,1%	14,8%	35,6%	0,1%
R54 Senility	2,1%	1,0%	45,4%	49,8%	0,6%
R55 Syncope and collapse	0,7%	0,1%	14,3%	31,2%	0,1%
R56 Convulsions, not elsewhere classified	0,5%	0,1%	22,5%	44,5%	0,1%
R59 Enlarged lymph nodes	0,2%	0,0%	8,3%	27,2%	0,0%
R60 Oedema, not elsewhere classified	0,1%	0,0%	14,8%	28,4%	0,0%
R62 Lack of expected normal physiological development	0,0%	0,0%	30,3%	33,6%	0,0%
R63 Symptoms and signs concerning food and fluid intake	0,5%	0,1%	21,3%	33,5%	0,1%
R68 Other general symptoms and signs	5,3%	0,1%	2,3%	33,9%	0,1%
R69 Unknown and unspecified causes of morbidity	13,6%	0,3%	1,9%	34,5%	0,3%
R70-R79 Abnormal findings on examination of blood, without diagnosis	1,1%	0,2%	14,3%	33,3%	0,1%
R87 Abnormal findings in specimens from female genital organs	0,6%	0,0%	6,5%	18,1%	0,0%
R95-R99 Ill-defined and unknown causes of mortality	0,0%				0,0%
S00-S09 Injuries to the head	2,0%	0,3%	13,9%	37,0%	0,2%
S10-S19 Injuries to the neck	0,1%	0,0%	21,4%	48,5%	0,0%

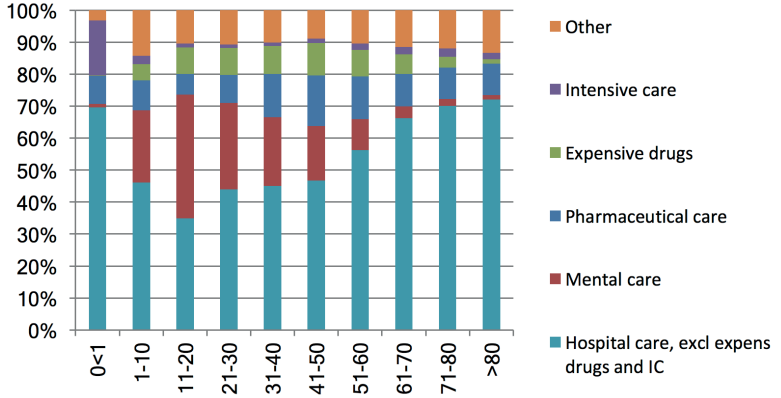
S20-S29 Injuries to the thorax	0,4%	0,2%	41,0%	49,7%	0,1%
S30-S39 Injuries to the abdomen, lower back, lumbar spine and pelvis	0,4%	0,2%	51,1%	40,1%	0,1%
S40-S49 Injuries to the shoulder and upper arm	1,0%	0,3%	35,3%	41,5%	0,2%
S50-S59 Injuries to the elbow and forearm	0,3%	0,1%	36,9%	41,5%	0,1%
S60-S69 Injuries to the wrist and hand	1,3%	0,2%	18,6%	31,5%	0,1%
S70-S79 Injuries to the hip and thigh	2,5%	2,2%	85,2%	62,4%	1,5%
S80-S89 Injuries to the knee and lower leg	0,7%	0,3%	46,6%	46,8%	0,2%
S90-S99 Injuries to the ankle and foot	1,1%	0,4%	34,8%	47,3%	0,2%
T00-T07 Injuries involving multiple body regions	0,9%	0,2%	22,4%	38,0%	0,1%
T08-T14 Injuries to unspecified part of trunk, limb or body region	5,1%	0,4%	8,3%	34,1%	0,3%
T15-T19 Effects of foreign body entering through natural orifice	0,1%	0,0%	8,5%	27,6%	0,0%
T20-T32 Burns and corrosions	0,1%	0,0%	28,3%	59,6%	0,0%
T33-T35 Frostbite	0,0%	0,0%	50,6%	2,7%	0,0%
T36-T50 Poisoning by drugs, medicaments and biological substances	0,6%	0,0%	7,4%	22,1%	0,0%
T51-T65 Toxic effects of substances chiefly nonmedicinal as to source	0,0%	0,0%	5,8%	18,4%	0,0%
T66-T78 Other and unspecified effects of external causes	0,4%	0,1%	17,7%	25,6%	0,0%
T79-T79 Certain early complications of trauma	0,0%	0,0%	33,8%	48,6%	0,0%
T80-T88 Complications of surgical and medical care, not elsewhere classified	1,2%	0,7%	54,8%	66,7%	0,5%
T90-T98 Sequelae of injuries, of poisoning and of other consequences of external causes	0,8%	0,2%	20,1%	43,1%	0,1%
Z00-Z13 Persons encountering health services for examination and investigation	3,8%	0,3%	8,6%	28,7%	0,2%
Z20-Z29 Persons with potential health hazards related to communicable diseases	0,0%	0,0%	4,1%	1,5%	0,0%
Z30-Z39 Persons encountering health services in circumstances related to reproduction	4,1%	0,7%	17,3%	46,4%	0,5%
Z40-Z54 Persons encountering health services for specific procedures and health care	10,6%	2,7%	25,7%	49,2%	1,8%
Z80-Z99 Persons with potential health hazards related to family and personal history and certain conditions influencing health status	9,6%	0,3%	3,0%	22,1%	0,5%

Appendix 2. Cross table describing patterns of health care use and demographics of top-2-5% beneficiaries

ICD10-chapter	Prevalence	% most cost incurring ICD10-chapter	Average age	Percentage dying	Percentage men	Average number of comor (ICD-chapter)	Average cost (*€1000)	Expensive drugs (*€1000)	Intensive care (*€1000)	Neoplasm (*€1000)	Mental and behavioural disorders (*€1000)	Diseases circulatory system (*€1000)	Diseases musculoskeletal system (*€1000)
Certain infectious and parasitic diseases	6.3%	3.2%	53.6	5.0%	70.8%	3.4	15	794	1,597	656	723	955	432
Neoplasms	27.4%	13.0%	65.9	14.8%	46.8%	3.2	17	28,980	6,602	224,741	2,120	6,267	2,829
Diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism	2.4%	0.6%	57.3	5.7%	46.6%	4.1	16	2,138	242	244	124	351	157
Endocrine, nutritional and metabolic diseases	20.2%	2.9%	42.3	2.0%	38.0%	3.4	15	6,748	1,122	518	1,051	955	662
Mental and behavioural disorders	26.6%	13.1%	41.1	1.0%	48.4%	2.6	16	4,216	2,067	1,597	217,204	3,339	3,022
Diseases of the nervous system	23.4%	2.8%	52.9	4.5%	44.7%	3.7	16	5,475	1,737	617	1,071	1,575	949
Diseases of the eye and adnexa	19.2%	1.5%	71.9	1.5%	42.4%	3.7	14	4,875	194	494	201	790	487
Diseases of the ear and mastoid process	4.9%	0.4%	48.4	1.1%	50.5%	3.4	14	654	70	72	176	116	88
Diseases of the circulatory system	40.3%	16.0%	69.3	8.1%	56.4%	3.7	17	5,085	17,064	5,430	3,416	267,652	4,555
Diseases of the respiratory system	19.2%	4.7%	66.6	13.0%	48.3%	3.9	16	4,054	6,835	2,491	1,582	4,232	1,431
Diseases of the digestive system	15.7%	6.1%	57.0	5.3%	44.4%	3.7	16	28,002	5,754	2,491	1,971	3,481	1,937
Diseases of the skin and subcutaneous tissue	8.2%	1.1%	58.7	3.5%	50.9%	3.5	15	8,449	217	271	250	566	336
Diseases of the musculoskeletal system and connective tissue	27.4%	13.4%	62.8	0.6%	35.1%	3.1	15	55,769	1,011	2,948	2,428	5,520	187,982
Diseases of the genitourinary system	17.0%	4.5%	59.3	4.8%	43.9%	3.8	15	3,545	1,993	2,235	1,259	2,411	1,445
Pregnancy, childbirth and the puerperium	4.0%	3.2%	31.0	0.0%	0.0%	2.7	13	884	564	178	1,661	413	348
Certain conditions originating in the perinatal period	0.0%	0.0%	0.0	0.0%	37.5%	2.6	18	0	26	0	0	0	0

Congenital malformations, deformations and chromosomal abnormalities	1.8%	0.5%	14.5	0.5%	54.7%	3.0	16	945	743	63	127	145	80
Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	28.2%	3.0%	66.4	15.1%	40.9%	4.0	16	2,223	2,390	1,266	1,354	2,338	1,066
Injury, poisoning and certain other consequences of external causes	15.9%	6.1%	67.8	9.2%	36.4%	3.5	16	1,957	4,388	1,497	2,283	3,554	3,483
Factors influencing health status and contact with health services	24.2%	3.9%	37.9	0.9%	40.8%	3.3	15	3,409	5,022	1,495	1,549	2,180	1,749

Appendix 3: Cost drivers per age group in top-2-5% beneficiaries



Appendix 4. Top-2-5% beneficiaries according to age group†

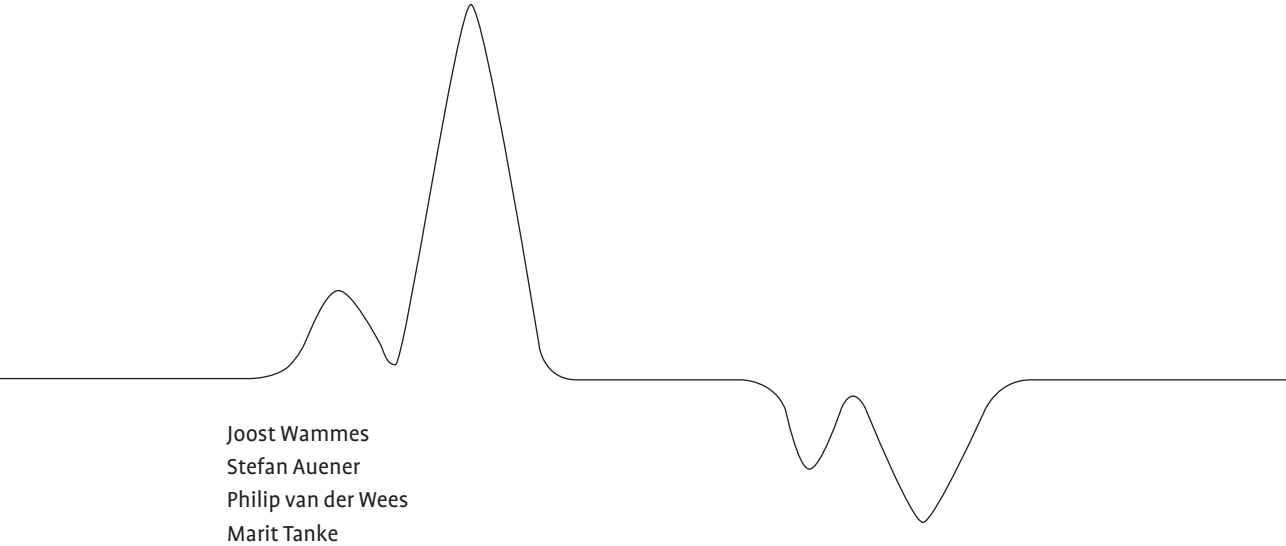
Age group	<1	1-10	11-20	21-30	31-40	41-50	51-60	61-70	71-80	>80
Number	1902	4311	6451	10612	14757	20971	26916	34047	33895	26964
Average per capita costs	€ 15,668	€ 15,704	€ 15,849	€ 14,900	€ 14,926	€ 15,648	€ 15,811	€ 15,971	€ 16,087	€ 16,038
Most important ICD-subchapters in terms of costs and share of total costs among the age group.	Factors influencing health status and contact with health services 33.8%	Mental and behavioral disorders 22.3%	Mental and behavioral disorders 35.5%	Mental and behavioral disorders 22.2%	Mental and behavioral disorders 17.6%	Mental and behavioral disorders 14.0%	Diseases of the circulatory system 10.2%	Neoplasms 13.8%	Diseases of the circulatory system 15.6%	Diseases of the circulatory system 17.0%
	Congenital malformations, deformations and chromosomal abnormalities 7.6%	Factors influencing health status and contact with health services 8.5%	Factors influencing health status and contact with health services 3.9%	Pregnancy, childbirth and the puerperium 12.9%	Pregnancy, childbirth and the puerperium 12.9%	Neoplasms 6.6%	Neoplasms 10.2%	Diseases of the circulatory system 13.7%	Neoplasms 12.2%	Injury, poisoning and certain other consequences of external causes 10.9%
	Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified 5.2%	Endocrine, nutritional and metabolic diseases 3.7%	Endocrine, nutritional and metabolic diseases 3.8%	Diseases of the digestive system 3.7%	Factors influencing health status and contact with health services 4.2%	Diseases of the circulatory system 6.1%	Diseases of the musculoskeletal system and connective tissue 8.2%	Diseases of the musculoskeletal system and connective tissue 11.0%	Diseases of the musculoskeletal system and connective tissue 10.8%	Neoplasms 7.2%

Diseases of the digestive system 3.3%	Congenital malformations, deformations and chromosomal abnormalities 3.6%	Injury, poisoning and certain other consequences of external causes 3.1%	Factors influencing health status and contact with health services 3.5%	Diseases of the musculoskeletal system and connective tissue 3.3%	Diseases of the musculoskeletal system and connective tissue 5.6%	Mental and behavioral disorders 8.0%	Diseases of the digestive system 3.4%	Injury, poisoning and certain other consequences of external causes 4.8%	Diseases of the musculoskeletal system and connective tissue 6.1%
Diseases of the respiratory system 2.7%	Diseases of the nervous system 2.7%	Diseases of the musculoskeletal system and connective tissue 2.9%	Diseases of the musculoskeletal system and connective tissue 2.3%	Diseases of the genitourinary system 3.2%	Factors influencing health status and contact with health services 2.6%	Diseases of the digestive system 3.5%	Injury, poisoning and certain other consequences of external causes 3.4%	Diseases of the respiratory system 3.7%	Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified 5.1%

† Total costs per ICD-chapter were summed per age group. In the table, the five ICD10-chapters with highest costs per age group are presented. I.e. among beneficiaries 1-10 years old, 22.3% of total costs were accounted for treatments for mental and behavioral disorders.

CHAPTER 8

Characteristics and healthcare utilization of patients with chronic heart failure and high costs; a longitudinal claim database analysis



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Abstract

Aims Costs are concentrated among so-called 'high-cost' patients. Many high-cost patients suffer from congestive heart failure and may be an interesting population to seek for quality improvement and cost containment. We determined the characteristics of patients with heart failure and high costs (top 1%, top 2-5% highest costs in perspective of the general population) and explored the longitudinal healthcare utilization and persistency of high costs.

Methods and results Longitudinal observational study using claims data from 2006-2014 in the Netherlands. We identified all patients that received a hospital treatment for chronic heart failure between 1 January 2008 and 31 December 2010. Our findings revealed that the difference in costs between top 1%, top 2-5% and bottom-95% patients with heart failure was mainly driven by hospital costs; and the top 1% group experienced a remarkable increase of mental health costs. More than 90% of the population incurred at least one top 5% year during follow-up, and 31.8% incurred at least one top 1% year. Top 1% and top 2-5% patients with heart failure differed from lower cost patients in their higher rate of chronic conditions, excessive polypharmacy, hospital admissions, and heart-related surgeries. Besides, top 1% patients were relatively young. Anemia, dementia, diseases of arteries veins and lymphatic vessels, influenza, and kidney failure were significantly associated with high costs. The end-of-life period was also predictive of top 1% and top 5% costs.

Conclusion Comprehensive and integrated efforts are needed to further improve quality of care and reduce unnecessary costs.

Introduction

It is known that healthcare costs are concentrated among so called 'high-cost patients' [1]. Although they receive substantial care from multiple sources, it is widely believed that many of these patients have critical unmet health care needs, and many receive unnecessary and ineffective care. In the US, about 40% of high-cost patients suffer from congestive heart failure (heart failure onwards) [2,3]. In earlier work, we found heart failure affected 12% of Dutch high-cost patients [1]. Repeated hospitalizations contribute to the high costs of patients with heart failure, and it is known that non-cardiovascular co-morbidities, a lack of multidisciplinary treatment, a lack of advance care planning, and a lack of guideline recommended care contribute to (preventable) hospitalizations [4,5]. Patients with heart failure may thus be exemplary for high-cost patients, and may be an interesting target population to seek for possible quality improvement and cost reduction.

A variety of interventions and programs have been developed to improve the quality and efficiency of care for patients with heart failure [6]. Heart failure clinics with specialised nurses have shown to improve clinical outcomes and reduce all cause and heart failure related readmissions [7]. For high-cost patients, studies have shown that the effectiveness and efficiency dramatically increase when interventions are targeted at the patients that are most likely to benefit [8]. It is thus of utmost important to acquire an in-depth understanding of the characteristics and healthcare utilization of patients with heart failure, and those with high longitudinal utilization in particular.

Little is known about the variety in characteristics and longitudinal healthcare utilization of patients with heart failure. Studies focusing on the costs of heart failure are scarce and the few that are available do not focus on high-cost patients within this population [9,10]. Furthermore, previous studies have focused on the prediction and prevention of hospital readmissions, the predominant cost driver of heart failure, and such studies are often limited by a short time horizon [11,12].

The overall objective of this study was to explore the characteristics and longitudinal healthcare utilization of patients with heart failure and high costs. We aimed to describe the characteristics of patients with heart failure and high costs, and to identify drivers of high costs. Furthermore, we aimed to study the longitudinal healthcare utilization of patients with heart failure, and to identify the persistency of high costs over time.

Methods

Design and context

The study was designed as a longitudinal observational study in routinely collected claims data. The study was situated in the curative health system in the Netherlands – a health insurance scheme based on the principles of managed competition that is governed by the Health Insurance Act. The system provides a wide range of services, including care provided by general practitioners, hospitals, and specialists; dental care through age 18; prescription drugs; physiotherapy through age 18; most mental care; medical aids and devices; maternity care; transportation and others. Voluntary complementary insurance benefits were excluded

from analysis, as were long-term care benefits that are covered under a separate scheme [13]. This study complies with the Declaration of Helsinki. No ethical approval is needed for this type of research in the Netherlands according to current legislation.

High-cost patients

Top-1% and top-5% of annual care utilization in perspective of the total population are widely used indicators for intensive utilization of health resources. We used the total beneficiary population (including beneficiaries without heart failure) to establish this characteristic per patient per year. The data were not available for the years before 2009. Therefore, the cut-off values for the top 1% and top 2-5% classes were extrapolated from subsequent years and used to determine top-1% and top-2-5% utilization.

Patient selection and data source

Data were drawn from the claims database of Zilveren Kruis, a health insurer currently covering 4.5 million beneficiaries who are primarily living in the central, eastern and western parts of the Netherlands. Detailed information about (a predecessor of) this database has been published in an earlier study [14].

We identified all patients that received an in- or outpatient hospital treatment for chronic heart failure between 1 January 2008 and 31 December 2010. We selected all patients with a claim containing specialism code 320 (cardiology) and diagnostic code 302 (chronic heart failure). We verified the correctness of this selection criterion with a cardiologist in our hospital. Analyses were limited to patients that were insured at the insurer during the entire study period or until death. Patients younger than 18, and patients who already received hospital treatment for heart failure before January 1 2008 were excluded.

Since our inclusion period covered a time horizon of three years, some patients received initial hospital treatment for heart failure in 2008 while others started heart failure treatment in 2009 or 2010. Therefore, years were recoded relative to the initial hospital treatment for heart failure. This enabled analysis of data relative to the first presentation of heart failure in hospitals (see figure 1).

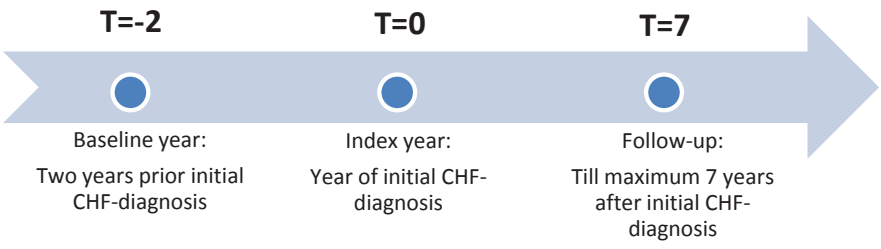


FIGURE 1 Timeline used to recode time relative to initial heart failure treatment.

Of each selected patient, all claims with a starting date between 1 January 2006 and 31 December 2014 were extracted. Pharmaceutical claims contained Anatomical Therapeutic Chemical (ATC) class codes, which were aggregated to ATC level 2. In addition, several beneficiary characteristics were obtained from the insurer's databases, including gender, date of birth, and date of death. Zip-codes (first four digits) were obtained to subsequently derive socio-economic status based on income estimates (appendix 1).

Variables

Literature was searched to identify factors known to affect the progression, prognosis and healthcare utilization of patients with heart failure [10,15,16]. Appendix 1 shows which variables were identified, and how the variables were operationalized in the present study. Polypharmacy was defined as receiving five or more prescription medications (ATC level 2) within a period of three months. We averaged this over a one year period to account for incidental medication [17]. Excessive polypharmacy was defined similarly for receiving at least ten prescription medicines.

We developed two measures to establish multimorbidity. Hospital DRGs in the Netherlands contain specialism and diagnosis codes, and we used these to categorize the claims according to ICD-10 (sub)chapters (e.g. ICD10-chapter IX: diseases of the circulatory system; and ICD10-subchapter I60-I69 cerebrovascular diseases). We summed all ICD-10 subchapters to establish a hospital-DRG-based multimorbidity measure. Second, we used pharmaceutical claims to establish chronic conditions based on a validated set of ATC-codes [18]. We summed al chronic conditions to establish a drug-based multimorbidity measure.

We used hospital claims to create dichotomous variables for heart related admissions and surgical interventions. Time since first hospital treatment for heart failure, previous healthcare expenditures and end-of-life period have all been identified as important cost drivers [19-21] and were included as well.

We combined chronic conditions derived from pharmaceutical claims and ICD-10 subchapters derived from hospital claims to establish dichotomous variables for specific conditions. Hospital claims and claims from specialized mental health institutions were combined to establish indicators for mental health care use.

Analyses

For each year, we determined the percentage of patients that incurred top-1% or top 2-5% costs. Descriptive analyses were performed to describe the characteristics of our study population at the index year (t=0). The analyses were performed separately for the hierarchical spending groups (top 1%, top 2-5%, and bottom 95% patients).

Longitudinal healthcare utilization and persistency of high costs

Descriptive analyses were used to analyze the longitudinal healthcare utilization per healthcare domain for the entire cohort, and per hierarchical spending group. The level of healthcare utilization during the index year (t=0) determined whether a patient was categorized to the top 1% , top 2-5%, or the bottom 95% subgroup for this analysis. In the following analyses all repeated measurements ($t \geq 0$) were our unit of analysis. We determined the percentage of top-1% and top-5% high-cost years, and identified the percentage of high-cost years that occurred consecutively.

Drivers of high costs

We used logistic generalized estimating equation (GEE) models to determine which factors were associated with high costs, while taking into account the clustering of repeated measurements within patients. We used GEE models with an exchangeable working correlation structure to account for this clustering [22]. In these analyses, repeated measurements (per year) were our unit of analysis; all follow-up years (t≥0) were analyzed. Our aim was to identify all factors significantly associated with high costs. Two types of dichotomous outcomes were analysed in separate models: 1) the top 1% as opposed to the lowest 99% cost years, and 2) the top 5% high cost years as opposed to the lowest 95% cost years. As independent variables we used all predictors, including demographics, disease specific variables, excessive polypharmacy, previous top-1%/top-2-5% healthcare utilization, heart related admission, heart related surgery, times since initial heart failure treatment in years, quarter of dying. All continuous variables were tested for the assumption of linearity and categorized if linearity could not be assumed. Backward selection was performed manually on basis of the type 3 significance tests ($p<0.05$), which is based on likelihood ratio statistics (PROC GENMOD in SAS). Associations were expressed as odds ratio's (OR) and 95% confidence intervals (95% CI). To determine the performance of the models, area under the curve (AUC) was assessed.

All analyzes were performed using SAS 9.4.

Results

Table 1 gives an overview of the repeated measurements (years) in our study. There were 25.372 unique patients with heart failure in our study. The percentage of patients that incurred top-1% or top-2-5% costs steadily increased until the index year (t=0). In the index year, the percentage incurring high costs was highest. From t=2 and onwards the percentage that incurred high costs levelled: 7% incurred top-1% costs, and 20% incurred top-2-5% costs.

TABLE 1 The percentage of top-1% and top-2-5% patients in each of the study years.

Year[1]	-4	-3	-2	-1	0	1	2	3	4	5	6
Total number of patients	8976	17327	25372	25372	25372	23714	21792	20133	18368	10859	4747
Top-1%	3%	4%	5%	7%	16%	9%	7%	7%	7%	7%	7%
Top-2-5%	13%	14%	16%	19%	34%	23%	22%	21%	20%	21%	20%
Bottom-95%	84%	82%	80%	74%	49%	68%	71%	72%	73%	72%	73%

[1] Since our inclusion period covered a time horizon of three years, some patients received initial hospital treatment for heart failure in 2008 while others started heart failure treatment in 2009 or 2010. Therefore, years were recoded relative to the initial hospital treatment for heart failure.

Characteristics during the index-year

Table 2 shows the characteristics for the three spending groups during the index year (t=0). More than half of the cohort incurred top 1% or top 2-5% costs. Top 1% patients were younger and the top 2-5% patients were older than those in the bottom 95%. Despite the difference (5.2 years) in age between the top 1% and the top 2-5% group, survival rates were similar. The rate of excessive polypharmacy was three times higher in the top-1% and top-2-5% groups than in the low-cost group. Most variability was observed in our multimorbidity measure based on hospital services: top 1%, top 2-5% and bottom 95% patients were treated for respectively 6.6, 4.9, and 2.9 ICD10-subchapters respectively. In addition, the three groups differed in their use of heart-related surgeries and admissions. The percentage admitted to the hospitals was four times higher in the top 1% group than in the bottom 95% group. Heart related surgeries were performed in 54% of top 1% patients. Not shown in the table: remaining top 1% patients differed in many aspects, most notably in their rate and intensity of mental health and pharmaceuticals use, and rates of chronic conditions and multimorbidity. They incurred 15.5% lower average costs.

TABLE 2 Characteristics of patients in hierarchical spending groups in the index year and survival after initial heart failure treatment.

	Bottom 95%	Top 2-5%	Top 1%
Demographics			
Percentage of total cohort	49.2%	34.5%	16.3%
Mean age in years	73.5	75.8	70.6
Gender = male	50%	48%	60%
Socioeconomic status = low	48%	51%	49%
Generic indicators of care needs			
Mean number of chronic conditions	3.1	4.2	4.4
Mean number of ICD-10 subchapters	2.9	4.9	6.6
Percentage polypharmacy (≥ 5 medications)	71%	90%	91%
Percentage excessive polypharmacy (≥ 10 medications)	11%	32%	36%
Percentage surgery (heart-related)	0.2%	10%	54%
Percentage admission (heart-related)	20%	65%	82%
Prevalence of conditions			
Anemia	6%	15%	20%
Cardiac arrest and arrhythmias	12%	20%	29%
Chronic lung disease	28%	41%	38%
Dementia	1%	4%	5%
Depression, anxiety and sleep disorders	12%	22%	25%
Diabetes	19%	30%	32%
Diseases of arteries, veins and lymphatic vessels	6%	12%	20%
Gout	5%	9%	10%

Hyperlipidimia		47%	51%	63%
Influenza, pneumonia or use of antibacterials		24%	46%	51%
Ischemic heart disease		9%	21%	36%
Kidney failure		2%	7%	13%
Neoplasms		14%	23%	25%
Pain		12%	24%	29%
Psychosis		2%	6%	7%
Thyroid disorders		7%	9%	8%
Valve disorders		4%	7%	12%
Adjustment and management of devices, cardiac rehabilitation and others		1%	3%	12%
Follow-up services after surgery		6%	12%	41%
Survival in years after the day of initial heart failure treatment	1	94%	85%	84%
	2	90%	76%	75%
	3	84%	67%	69%
	4	78%	59%	62%
	5	72%	51%	54%

Longitudinal healthcare utilization

Figure 2 shows the average total costs over time for the full cohort of patients, and separately for survivors (those alive at the latest year with cost data). The overall patterns of utilization were similar. Highest average costs were found during the index year. The average cost per patient increased between t=-2 to t=0, and this increase was mainly driven by increasing hospital costs. After the year of initial heart failure treatment, costs quickly declined and stabilized at a level that was significantly higher than in the years prior initial heart failure treatment. Hospital costs were the predominant cost drivers in all years, followed by pharmaceutical costs. Not shown in the figure is that at any individual year, decedents incurred 90% higher costs than remaining patients in that year. The overall average costs in figure 2 are similar because in each year ≈10% of patients die.

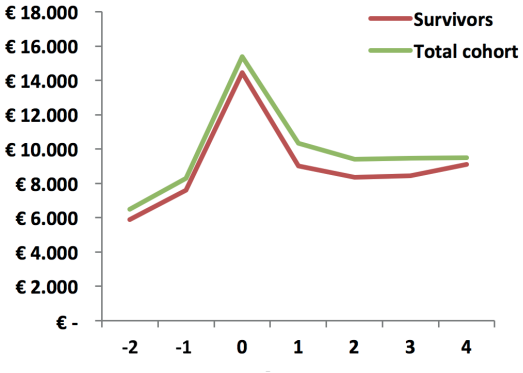


FIGURE 2 Average costs for survivors and the total cohort.

Appendix 2 shows the same healthcare utilization patterns, but for the three hierarchical spending groups separately. Groups were based on healthcare utilization during the index year. During the index year, average costs in the top 1% group (€ 48.120) were ten times as high compared to the bottom 95% group (€ 4.627). Top-1% incurred higher costs in each healthcare domain at any moment. This difference in total costs between the hierarchical spending groups was mainly driven by differences in hospital costs. The top 1% group experienced a remarkable increase of mental health care costs during the index year.

Persistency of high costs

Figure 3a and 3b show the persistency of high costs after first heart failure treatment (t≥0) for top 5% and top 1% utilization respectively. The height of the bars (y-axis) indicate the percentage of the cohort incurring a certain number of high-cost years (x-axis). Colour saturation shows the proportion of high-cost years which occurred consecutively. While more than 90% of the population incurred at least one top 5% year during follow-up, only 31.8% incurred at least one top 1% year. Furthermore, 57.0% incurred multiple top 5% years whereas only 8.6% incurred multiple top 1% years. In addition, top 5% years were more frequently consecutive than top 1% years.

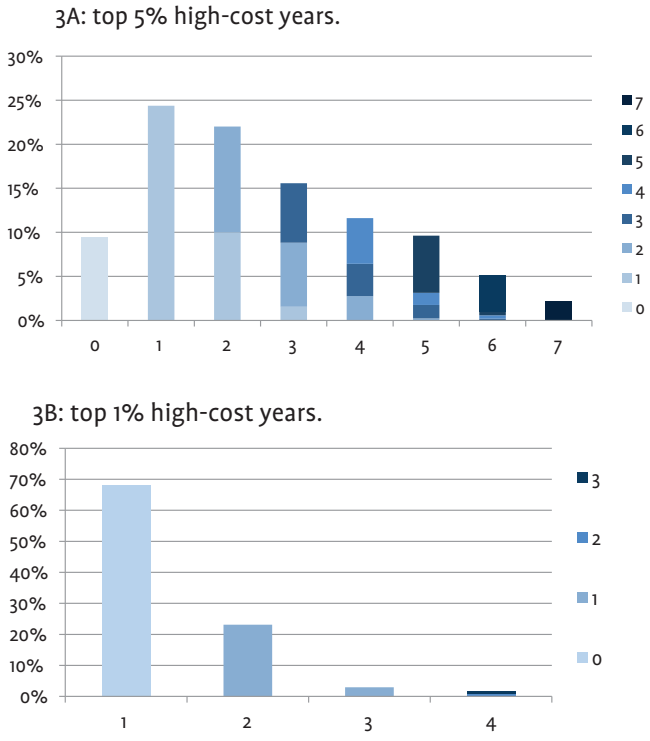


FIGURE 3 Frequency and persistency of high-cost years during follow-up period (t≥0). For example, figure 3a shows that 22% of the cohort had two top 5% years: 12% experienced two consecutive top 5% years, and 10% experienced two non-consecutive top 5% years.

Drivers of high costs

GEEs were performed for top 1% or top 5% high-cost years compared to bottom 99% and bottom 95% years, respectively. As explained above, all repeated measurements (t≥0) were our unit of analysis. Of the 125.166 follow-up years included in this study, 11.483 (9.2%) and 30.056 (24.0%) were top 1% and top 2-5% high-cost years, respectively.

Table 3 shows the estimated odds ratios (OR) for our final models. Younger groups were more likely to incur top 1% costs. Excessive polypharmacy, high costs in the previous year, and end-of-life periods were all predictive of top 1% and top 5% costs. Heart related surgeries and heart related admissions showed highest OR's. In year one and two after initial heart failure treatment the odds of high costs were decreased, and in the following years the odds of high costs increased. Influenza was a specific disease with a high OR for high costs as well as a high prevalence among high-cost patients (see table 2).

TABLE 3 Odds Ratios for high cost years derived from GEE estimates: Diseases specific model.

		Top 1% year	Top 5% year
Variables		**OR (95% CI)**	**OR (95% CI)**
Gender male ref=female		1.17 (1.11-1.24)	NS
Age	60-69	0.86 (0.78-0.94)	1.00 (0.93-1.07)
Ref=18-59	70-79	0.71 (0.65-0.78)	1.05 (0.98-1.12)
	80-89	0.41 (0.38-0.46)	0.93 (0.87-0.99)
	≥90	0.23 (0.20-0.27)	0.73 (0.67-0.79)
Socioeconomic status	Average	NS	0.93 (0.89-0.98)
Ref = high	Low	NS	0.99 (0.95-1.03)
Excessive polypharmacy		1.56 (1.47-1.66)	1.95 (1.88-2.03)
Heart related surgery		22.00 (20.08-24.09)	65.08 (51.16-82.80)
Heart related admission		2.38 (2.22-2.55)	6.77 (6.45-7.11)
Time since heart failure treatment in years	1	0.57 (0.52-0.61)	0.56 (0.52-0.59)
Ref= 0 (year of initial hospital treatment)	2	0.80 (0.75-0.87)	0.79 (0.75-0.83)
	3	1.16 (1.07-1.25)	0.97 (0.92-1.01)
	4	1.63 (1.50-1.77)	1.15 (1.09-1.21)
	5	1.62 (1.47-1.78)	1.19 (1.12-1.27)
Quarter of dying	1	0.37 (0.28-0.48)	0.41 (0.36-0.47)
Ref=0 (survived entire year)	2	0.98 (0.83-1.16)	1.23 (1.10-1.38)
	3	1.83 (1.59-2.10)	2.33 (2.08-2.60)
	4	2.82 (2.52-3.15)	3.93 (3.55-4.35)
Top 1% in the previous year		3.51 (3.21-3.84)	3.04 (2.87-3.22)
Top 2-5% in the previous year		1.76 (1.67-1.86)	1.74 (1.67-1.81)
Disease specifc variables			
Anemia		1.66 (1.55-1.77)	1.94 (1.85-2.04)
Cardiac arrest and arrythmias		0.91 (0.85-0.98)	NS
Chronic lung disease		NS	1.38 (1.33-1.43)
Dementia		1.90 (1.66-2.18)	2.27 (2.03-2.52)
Depression, anxiety and sleep disorders		1.34 (1.25-1.43)	1.44 (1.37-1.51)
Diabetes		1.13 (1.07-1.20)	1.40 (1.35-1.45)

Diseases of arteries, veins and lymphatic vessels	2.26 (1.09-2.46)	1.93 (1.80-2.06)
Gout	1.15 (1.06-1.24)	1.21 (1.15-1.28)
Influenza, pneumonia or use of antibacterials	1.81 (1.72-1.90)	2.04 (1.97-2.10)
Ischemic heart disease	0.74 (0.68-0.81)	NS
Kidney failure	2.11 (1.90-2.34)	2.10 (1.93-2.28)
Neoplasms	1.70 (1.59-1.82)	2.00 (1.91-2.10)
Pain	1.60 (1.51-1.69)	1.87 (1.80-1.94)
Psychosis	1.32 (1.19-1.47)	1.41 (1.31-1.53)
Valve disorders	1.44 (1.31-1.60)	NS
Adjustment and management of devices, cardiac rehabilitation, and others	1.26 (1.10- 1.44)	1.33 (1.12-1.58)
Follow-up services after surgery	NS	0.71 (0.67-0.76)
AUC	0.87	0.85

NS: Some variables were excluded in the backward selection process in the model for top-1% and not for the top-5%, and vice versa.

Discussion

In this study, we explored the longitudinal healthcare utilization and the persistency of high costs in patients with heart failure. Furthermore, we determined the characteristics of patients with heart failure and high costs, and identified drivers of high costs. Our findings revealed that the difference in costs between the three groups was mainly driven by hospital costs. In addition, the top 1% group experienced a remarkable increase of mental health costs during the index year. More than 90% of the population incurred at least one top 5% year during follow-up, and 31.8% incurred at least one top 1% year. Top 5% years were more frequently consecutive than top 1% years. Top 1% and top 2-5% patients with heart failure differed from lower cost patients in their higher rate of chronic conditions, excessive polypharmacy, hospital admissions, and heart-related surgeries. Besides, top 1% patients were relatively young and elder patients were less likely to incur a top 1% year. Several of the disease specific variables showed significant OR's for high costs, including anemia, dementia, diseases of arteries veins and lymphatic vessels, influenza, and kidney failure. The end-of-life period was also predictive of top 1% and top 5% costs. These results provide necessary information for further increasing quality of care and reducing costs for patients with heart failure.

Strengths and limitations

To our knowledge, this is the first longitudinal study focusing on high-cost patients within a population of patients with heart failure. By using administrative data from our country's largest health insurer, we created a large set of variables that covered demographic characteristics, chronic conditions, hospital treatments and mental health utilization. This

allowed us to assess which characteristics were particularly associated with high costs. Due to having data of multiple consecutive years, we were also able to explore the longitudinal healthcare utilization and persistency of high costs. One limitation was our lack of clinical data, data of long-term care, and individual patient data of quality of care. Such data could facilitate a deeper understanding of healthcare utilization, care needs and opportunities to intervene in patients with heart failure.

Reflection on our findings

Our findings generally align with prior research, which supports the generalizability of our findings. The prevalence of most comorbid conditions such as chronic lung diseases, diabetes, anemia and depression were similar to previous studies [15,16,23-25], as was rate of mortality [26]. We found that the odds of high costs decreased in the two years following initial heart failure treatment, and increased in the years thereafter. This corresponds with the progressive nature of heart failure and associated increase of healthcare needs [10, 21]. The relatively high costs at initial diagnosis are surprising, and may reflect extensive diagnostic trajectories or time for the treatment to take effect.

We were the first to explore the frequency and persistency of high costs in patients with heart failure. Our findings indicate that top-1% utilization predominantly occurs incidentally and among less than a third of patients with heart failure, whereas almost all patients with heart failure experience at least one top 5% year, and more than half experience two or more top 5% years. Our breakdown of characteristics and cost drivers revealed the most important cost drivers in patients with heart failure. Heart-related surgeries contributed to the incidental high costs in 54% of top 1% patients, and the costs of the remaining top 1% patients were driven by mental health and pharmaceuticals use, and rates of chronic conditions and multimorbidity. The high frequency and persistency of top 5% utilization point to the well-known fact that heart failure is a devastating disease with severe symptoms, which is often accompanied by many comorbidities and low quality of life, which requires intensive medical treatment.

Our work contributes to existing literature because of our extensive inclusion of potential drivers for high costs. Wammes et al. and Joynt et al. argued that expensive procedures may be a more significant cost driver in high-cost patients than avoidable hospitalizations [1,3]. Our results confirm that procedures are important cost drivers in patients with heart failure. Besides, our findings point to a select set of key cost drivers. Such drivers include chronic conditions and multimorbidity, excessive polypharmacy, and mental healthcare needs. Furthermore, we found that decedents incurred 90% higher costs in the year they died. Reducing end-of-life expenditures are important targets for intervention. However, the benefits of interventions aimed at longer term drivers of high costs may be of more importance if one seeks for additional value and efficiency for these patients.

Policy and research implications

Many initiatives to stimulate value and efficiency of care among patients with heart failure primarily concern reducing heart failure related re-admissions. For example, disease management programs at heart failure clinics have shown to improve patient well-being, reduce both hospitalizations and mortality, and may even save costs. Key ingredients of such

programs are guideline adherence and the integration and coordination of multidisciplinary heart failure treatment across the continuum of care, which includes treatment by specialized heart failure cardiologists and specialized heart failure nurses, adequate post-discharge planning, and advance care planning in advanced heart failure [27].

Our findings revealed a range of drivers for high costs that may be beyond the scope of such initiatives. The scope of care improvement programs may be widened to include also the treatment of common co-morbidities. Moreover, it is widely known that mental care needs are underestimated in heart failure patients and may be underserved in current health systems, and timed treatment, or tailored treatment for heart failure induced depression, might have prevented the high mental care expenditures we observed.

Furthermore, identified indicators may reflect overuse of care. The optimal indication criteria for surgical interventions tend to evolve in time, and in the Netherlands there is a nascent trend towards operating less in (frail) elderly. Unnecessary transaortic valve replacements were reduced through a multidisciplinary approach [28]. Research of medical practice variation has identified unwarranted variation in a range of services [29]. Especially near the end of life, patient preferences vary substantially and shared-decision making is warranted [30].

This study used administrative data from the perspective of patients with heart failure and high costs, in order to inform policy and practice. Inclusion of clinical data, patient-reported outcome measures and of quality of care might further improve the validity and actionability of our findings. For example through identification of organisational characteristics (at hospital or health system level) or processes that are associated with costs, outcomes of care, and/or unwarranted variation of care. In addition, further research may be needed to discern preventable spending from high-value spending in patients with heart failure, and further research is needed to study the effects of organisational factors and medical practice variation towards high costs in patients with heart failure.

In conclusion, our study has addressed persistently high costs and drivers of high costs in patients with heart failure. Comprehensive and integrated efforts are needed to further improve quality of care and reduce unnecessary costs.

References

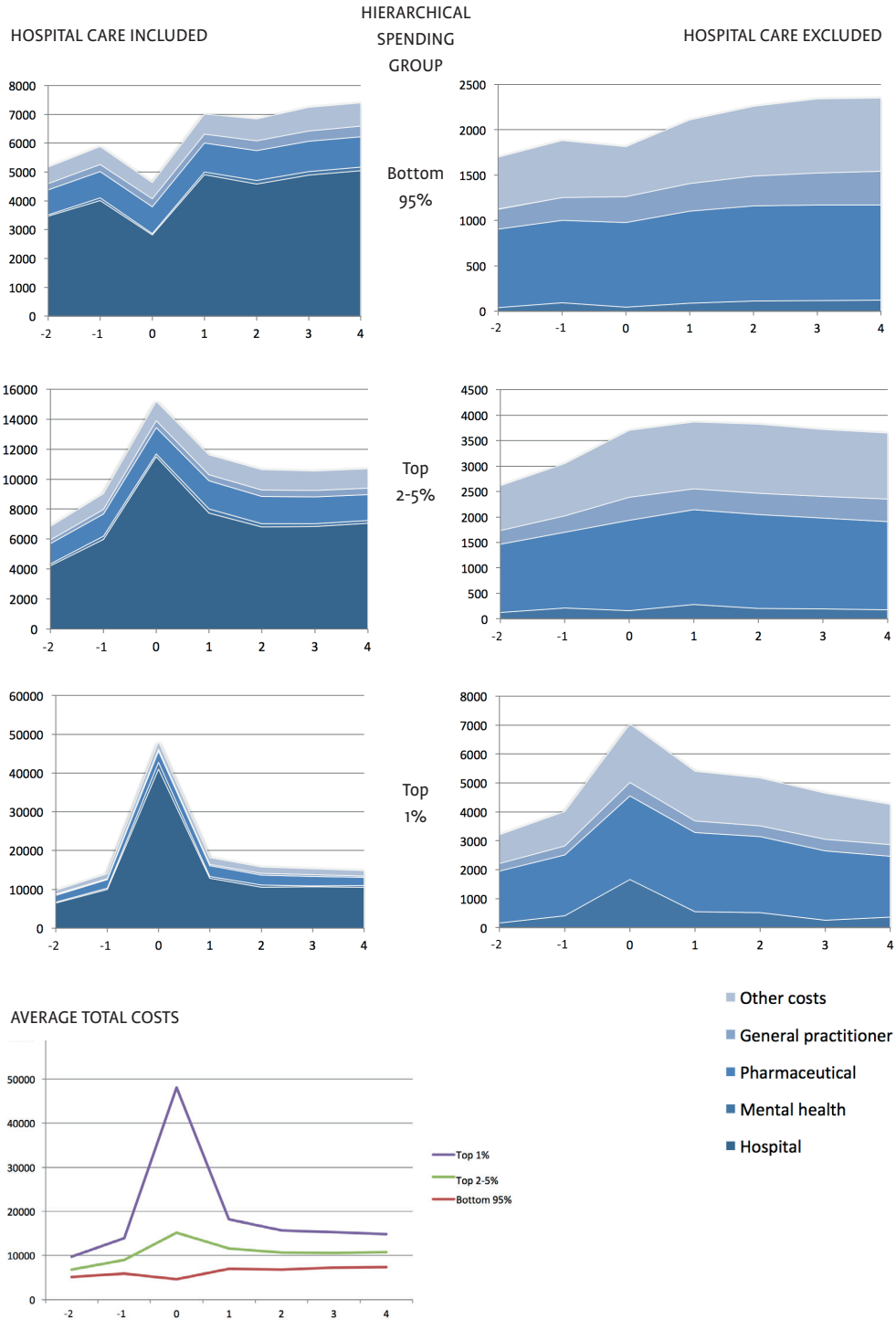
1 Wammes JJG, Tanke M, Jonkers W, Westert GP, Van der Wees P, Jeurissen PP. Characteristics and healthcare utilisation patterns of high-cost beneficiaries in the Netherlands: a cross-sectional claims database study. BMJ open. 2017;7(11):e017775.
2 Figueroa JF, Frakt AB, Lyon ZM, Zhou X, Jha AK. Characteristics and spending patterns of high cost, non-elderly adults in Massachusetts. Healthcare (Amsterdam, Netherlands). 2017.
3 Joynt KE, Gawande AA, Orav EJ, Jha AK. Contribution of preventable acute care spending to total spending for high-cost Medicare patients. Jama. 2013;309(24):2572-8.
4 Saito M, Negishi K, Marwick TH. Meta-Analysis of Risks for Short-Term Readmission in Patients With Heart Failure. Am J Cardiol. 2016;117(4):626-32.
5 Gheorghiade M, Vaduganathan M, Fonarow GC, Bonow RO. Rehospitalization for heart failure: problems and perspectives. J Am Coll Cardiol. 2013;61(4):391-403.
6 Roccaforte R, Demers C, Baldassarre F, K Teo K, Yusuf S. Effectiveness of comprehensive disease management programmes in improving clinical outcomes in heart failure patients. A meta-analysis. European journal of heart failure. 2005;7(7):1133-44.
7 Takeda A, Taylor SJ, Taylor RS, Khan F, Krum H, Underwood M. Clinical service organisation for heart failure. Cochrane Database Syst Rev. 2012(9):CD002752.
8 Brown RS, Peikes D, Peterson G, Schore J, Razafindrakoto CM. Six features of Medicare coordinated care demonstration programs that cut hospital admissions of high-risk patients. Health affairs. 2012;31(6):1156-66.
9 Berry C, Murdoch DR, McMurray JJ. Economics of chronic heart failure. European journal of heart failure. 2001;3(3):283-91.
10 Smith DH, Johnson ES, Blough DK, Thorp ML, Yang X, Petrik AF, et al. Predicting costs of care in heart failure patients. BMC health services research. 2012;12(1):434.
11 Au AG, McAlister FA, Bakal JA, Ezekowitz J, Kaul P, van Walraven C. Predicting the risk of unplanned readmission or death within 30 days of discharge after a heart failure hospitalization. American heart journal. 2012;164(3):365-72.
12 Desai AS, Stevenson LW. Rehospitalization for heart failure. Circulation. 2012;126(4):501-6.
13 Kroneman M, Boerma W, van den Berg M, Groenewegen P, de Jong J, van Ginneken E. The Netherlands: health system review.2016.
14 Smeets HM, de Wit NJ, Hoes AW. Routine health insurance data for scientific research: potential and limitations of the Agis Health Database. J Clin Epidemiol. 2011;64(4):424-30.
15 Ezekowitz JA, McAlister FA, Armstrong PW. Anemia is common in heart failure and is associated with poor outcomes: insights from a cohort of 12 065 patients with new-onset heart failure. Circulation. 2003;107(2):223-5.
16 Chamberlain AM, Sauver JLS, Gerber Y, Manemann SM, Boyd CM, Dunlay SM, et al. Multimorbidity in heart failure: a community perspective. The American journal of medicine. 2015;128(1):38-45.

17 Bjerrum L, Rosholm J, Hallas J, Kragstrup J. Methods for estimating the occurrence of polypharmacy by means of a prescription database. European journal of clinical pharmacology. 1997;53(1):7-11.
18 Huber CA, Szucs TD, Rapold R, Reich O. Identifying patients with chronic conditions using pharmacy data in Switzerland: an updated mapping approach to the classification of medications. BMC public health. 2013;13(1):1030.
19 Levenson JW, McCarthy EP, Lynn J, Davis RB, Phillips RS. The last six months of life for patients with congestive heart failure. Journal of the American Geriatrics Society. 2000;48(S1).
20 Tamang S, Milstein A, Sørensen HT, Pedersen L, Mackey L, Betterton J-R, et al. Predicting patient 'cost blooms' in Denmark: a longitudinal population-based study. BMJ open. 2017;7(1):e011580.
21 Dunlay SM, Shah ND, Shi Q, Morlan B, VanHouten H, Long KH, et al. Lifetime costs of medical care after heart failure diagnosis. Circulation: Cardiovascular Quality and Outcomes. 2011;4(1):68-75.
22 Hanley JA, Negassa A, Edwardes MD, Forrester JE. Statistical analysis of correlated data using generalized estimating equations: an orientation. American journal of epidemiology. 2003;157(4):364-75.
23 Cohen-Solal A, Beauvais F, Logeart D. Heart failure and diabetes mellitus: epidemiology and management of an alarming association. Journal of cardiac failure. 2008;14(7):615-25.
24 Hawkins NM, Petrie MC, Jhund PS, Chalmers GW, Dunn FG, McMurray JJ. Heart failure and chronic obstructive pulmonary disease: diagnostic pitfalls and epidemiology. Eur J Heart Fail. 2009;11(2):130-9.
25 Rutledge T, Reis VA, Linke SE, Greenberg BH, Mills PJ. Depression in heart failure: a meta-analytic review of prevalence, intervention effects, and associations with clinical outcomes. Journal of the American college of Cardiology. 2006;48(8):1527-37.
26 Gottdiener JS, McClelland RL, Marshall R, Shemanski L, Furberg CD, Kitzman DW, et al. Outcome of congestive heart failure in elderly persons: influence of left ventricular systolic function. The Cardiovascular Health Study. Annals of internal medicine. 2002;137(8):631-9.
27 Moertl D. Disease management programs in heart failure: half a century of an unmet need. Wien Klin Wochenschr. 2017;129(23-24):861-3.
28 Schoon Y, de Boer M, Olde Rikkert M. Niet elke oudere heeft baat bij nieuwe hartklep. 2015.
29 Roth GA, Brown J, Malenka DJ. Medical Practice Variations in Heart Failure. In: Johnson A, Stukel T, editors. Medical Practice Variations. New York: Springer-Verlag New York Inc.; 2016.
30 Dev S, Abernethy AP, Rogers JG, O'Connor CM. Preferences of people with advanced heart failure-a structured narrative literature review to inform decision making in the palliative care setting. Am Heart J. 2012;164(3):313-9 e5.

Appendix 1. Definition of variables

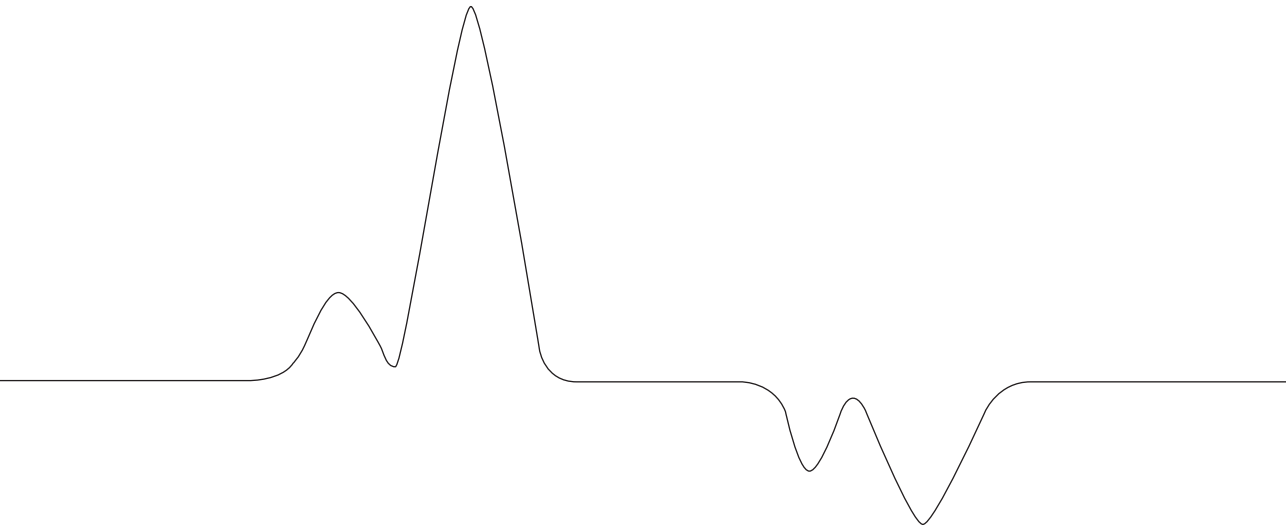
Variable	Data source/definition	
Demographics		
Sex	Insurance file	
Age	Insurance file	
Date of death	Insurance file	
Socioeconomic status	Derived from postal ZIP-code (first four digits)	
Generic characteristics		
Surgery by cardiologist	Specialism code 320, treatment code 11-36	
Surgery by cardiothoracic surgeon	Specialism code 328, diagnostic code 2210-2940	
Cardiology related admission	Specialism code 320, setting code 3	
Number of ICD10-subchapters	Identified from hospital claims	
Polypharmacy	≥5 prescription drugs ATC level 2	
Excessive polypharmacy	≥10 prescription drugs ATC level 2	
High costs in previous year	Top 1%, top 2-5% in the previous year	
Time since initial heart failure treatment	Initial heart failure treatment in hospital, identified from hospital claims	
Disease specific indicators	Hospital DRG-based (ICD-10 subchapter)	Medication-based (ATC code)
Anemia	D50-D59	B03Ax
Cardiac arrest and arrythmias	I44-I49	-
Chronic lung disease	J40-J47	R03Ax or R03Bx
Dementia	F00-F09	N06Dx
Depression, anxiety and sleep disorders	F30-F48	N05Bx or N05Cx or N06Ax
Diabetes	E10-E14	A10Ax or A10Bx or A10X
Diseases of arteries, veins and lymphatic vessels	I70-I89	-
Gout	-	M04A x
Heart failure	I50	-
Hyperlipidimia	-	C10x
Influenza, pneumonia or use of antibacterials	J09-J18	J01Cx or J01Mx
Ischemic heart disease	I20-I25	-
Kidney failure	N17-N19	-
Neoplasms	C00-D49	L01x
Pain	-	N02Ax or N02Bx
Psychosis	F20-F29	N05Ax
Thyroid disorders	E00-E07	H03x
Valve disorders	I34-I39	-
Adjusting of pacemakers, cardiac rehabilitation and other treatments coverder by ICD10-subchapter Z40-Z54 and performed by a cardiologist	Z40-Z54 and specialism code 320	-
Heart transplants and other treatments covered by ICD10-subchapter Z80-Z99 and performed by a cardiologist	Z80-Z99 and specialism code 320	-

Appendix 2: Cost trajectories per healthcare sector per hierarchical spending group. Costs in € (y-axis) over time (x-axis)



CHAPTER 9

General discussion



The studies in this thesis explored two approaches for reducing unnecessary and possibly harmful care – so-called low-value services – while simultaneously improving outcomes of care, and reducing spending. As such, this thesis was aimed at improving the fiscal sustainability of healthcare. The first approach aimed to track down unnecessary care and to identify the determinants of unnecessary care provision in Dutch healthcare; in order to effectively reduce unnecessary spending. The second approach encompassed an exploration of the characteristics and utilization of high-cost patients; the sickest patients who are in heaviest need for care, but who are at highest risk to receive suboptimal treatment and unnecessary care. Our research questions were:

1 What are opportunities for cost-reduction through reduction of low-value services in the Netherlands?
2 What are the characteristics and healthcare utilization of high-cost patients and what strategies do likely improve high-cost patients' care and reduce costs?

This chapter starts by providing answers to the research questions outlined above. Next, several methodological considerations and lessons for future research are discussed. Furthermore, the implications of our research for future policy and research are discussed. Finally, an overall conclusion will be given.

This thesis was situated in the Netherlands. Chapter two gives an overview of the current health system of the Netherlands.

Main findings

Opportunities for cost-reduction through reduction of low-value services

We broke down our first research question into four sub-questions, which will be discussed below.

A *In which healthcare domains does low-value care typically prevail?*

This thesis was (partly) inspired by a remarkable finding in the 2012 International Health Policy (IHP) survey: more than half (57%) of the surveyed Dutch general practitioners (GPs) perceived that Dutch patients receive (much) too much medical care. *Chapter three* was aimed at understanding this figure, and to track the amount of unnecessary care across healthcare domains and care types through an exploratory survey among Dutch GPs. The surveyed GPs pointed to a remarkably consistent pattern of unnecessary service use: too much care is delivered in private clinics, at GP cooperatives, in hospitals, and by general practitioners themselves. The consensus was that patients receive too much diagnostic care, medical treatments, as well as too much monitoring and follow-up.

B W*hat are the main determinants for low-value care provision?*

In *chapter three* we also identified a range of determinants that are associated with too much care provision, both in general practices as well as in other healthcare domains, including hospitals. The chapter demonstrated that the GPs' demand-satisfying attitude and the increased availability of diagnostic facilities most saliently contribute to the provision of

perceived excess care in general practices. Patients tend to experience healthcare as a right to receive, and many GPs are willing to comply with the wishes of the patients in order to maintain their relationship. In addition, GPs may be willing to provide unnecessary care, in order prevent much more unnecessary care elsewhere (in hospitals). Moreover, our analysis pointed to a range of financial incentives that were associated with unnecessary care provision, both in general practices as in hospitals. For example, funding gaps between primary care and hospitals impede cooperation and coordination, and this provokes unnecessary care.

Together, our findings indicate that Dutch GPs have a clear view on which of the treatments their patients receive may be unnecessary. Besides, our findings show GPs are willing to reduce unnecessary spending, but that the system is misaligned, and that the GPs' ability to further improve the financial sustainability of health care may be limited. The new covenant for primary care has addressed some of these concerns. For example, there will be more time for pro-active, person-centred and integrated care for frail elderly; relevant stakeholders intend to establish local cooperation agreements to stimulate integrated care; and there will be further investments in information technology [1].

C *How to identify low-value services from clinical practice guidelines?*

We developed a standardized approach to identify low-value services from medical practice guidelines. *Chapter four* describes the development of the Dutch do-not-do list (Beter-Niet-Doen lijst). On the basis of a shortlist of search terms, a total of 1366 lower value services was found in 193 Dutch hospital guidelines. Of the lower value services 30% covered diagnostics, 29% related to surgical and medical treatment without drugs and 39% related to drug treatment. The majority (77%) of all low-value services was on care that should not be offered at all, whereas the other 23% recommended on care that should not be offered routinely.

It has often been said that due to a lack of clinical evidence, it is simply not known which services are of high-value, and which services should be considered low-value. This is partly true, it is known that for 50% of the treatments the effectiveness is unknown [2]. However, our findings show that for many services there is broad consensus in medical practice guidelines that the use of these low-value services should be very much reduced, if not totally abolished.

D *How do Dutch healthcare providers deal with the entry of low-value and cost-ineffective services and what policy might improve this?*

In *chapter five* we studied how cost-increasing services have entered Dutch hospitals and what services were displaced to accommodate the entry of these services. We interviewed 84 professionals with various roles and responsibilities (practitioners, department chairs, board of directors, insurers, and others). Our findings show that it is difficult to identify the services that are displaced to accommodate the cost-increasing health technologies; limited transparency in the flow of funds within a hospital contributed to this. Besides, we found that the entry of new innovations and cost-containment are two parallel processes that are not causally linked. The way of financing is pivotal in displacement in the Netherlands. The budget pressure of expensive drugs seems to be linked to horizontal reallocation across

departments, whereas the budget pressure of remaining services seems to be linked to vertical reallocation within departments or divisions. Hospitals have reacted to budget pressures primarily through a narrowing in the portfolio of their services, and a range of other efficiency measures. The board of directors is central in these processes, insurers are involved only to a limited extent at a high level of healthcare purchasing. Capacity (constraints) and financing are pivotal in understanding displacement effects.

Characteristics and healthcare utilization of high-cost patients

This part of the thesis was inspired upon the well-known fact that healthcare costs are heavily skewed towards a small share of high-cost patients. It is thus necessary to acquire an in-depth understanding of the characteristics, healthcare utilization and other factors that drive the costs of high-cost beneficiaries.

Chapter six presented our systematic review of high-cost patients' characteristics and healthcare utilization, and chapter seven presented our Dutch claim database analysis on the same issue. Both studies showed that high-cost patients are overwhelmingly characterized by multiple (chronic) conditions, and that many high-cost patients suffer from mental and behavioural disorders. Our review highlighted that many health system characteristics may contribute to high costs, and that 'preventable' spending was estimated at maximally ten percent of spending. Furthermore, a considerable share (approximately 40%) of high-cost patients persistently incurs high costs over the years. In addition, high-cost patients are more likely to die, and decedents are more likely to incur high-costs. However, no more than 30% of high-cost patients are in their last year of life. Besides, we identified a range of diverging cost drivers across payers and countries, which suggests that tailored approaches are needed for improving care and reducing costs. Our Dutch study showed that expensive services (expensive drugs, ICU treatment, dialysis, transplant care, DRG > €30,000) contributed to high costs in about a third of Dutch top 1% patients, and in less than ten percent of top 2-5% patients. Besides, high-cost patients were overwhelmingly treated for diseases of circulatory system, neoplasms, and mental disorders. Finally, in both studies we found that elderly are generally overrepresented in high-cost patients; but that more than halve of high-cost patients are younger than 65 of age, and in the Netherlands the average costs sharply declined with age within the top 1%.

We chose patients with heart failure to further study (persistency of high) utilization in high-cost patients. *Chapter eight* showed that more than 90% of patients with heart failure incurred at least one top 5% year, and 32% incurred at least one top 1% year. Besides, top-1% utilization predominantly occurs incidentally, whereas more than half experience two or more top 5% years and the majority of these top 5% years were incurred consecutively. Patients with heart failure and top 1% and top 2-5% utilization differed from others in their higher rate of chronic conditions and multimorbidity, excessive polypharmacy, hospital admissions, and heart-related surgeries. In addition, the top 1% group experienced a remarkable increase of mental health care costs during the initial year with heart failure.

One important empirical question is whether low-value services (first approach) or 'preventable spending' is concentrated among high-cost patients (second approach). In chapter six we found that this was the case: Figueroa et al. found that 4.8% of US Medicare spending was preventable, and that high-cost patients accounted for 73.8% of preventable

spending [3]. Similarly, McWilliams and Schwartz found that the 17% highest-risk patients received twice as many low-value services (31 low-value services, detected in claims) as lower-risk patients. However, their argument was that patient-focused strategies are not directly targeted towards low-value services, and as such must be substantially more effective than system-focused efforts intended to reduce low-value services, in order to achieve an equal number in the total number reduced [4]. However, this reasoning is very much dependent on what is considered of low-value.

Methodological considerations

This thesis is timely in an era of increasing healthcare costs, and explores two novel approaches for stimulating quality of healthcare while simultaneously reducing costs. An interdisciplinary approach was taken through a variety of research methods, including survey research, document analysis, a qualitative interview study, a systematic review, and claim database analyses. The specific limitations of each study have already been discussed in each of the chapters. Below, general methodological considerations are presented.

One main limitation of this thesis is that it lacks a direct estimation of the prevalence of low-value or unnecessary care in Dutch healthcare. This is partly due to a lack of data with sufficient clinical detail. Furthermore, as we have shown in chapter one, there is a lack of agreement on how to discern low-value from high-value care, which may be partly be a normative rather than scientific question. Moreover, there is a general lack of evidence of the value of most of medical services. Additionally, the value of care may very much depend upon the preferences of individual patients, and such preferences are not always noted in electronic health records, and never available in claim databases. Finally, there is a general lack of data on the outcomes of care, and such data may be needed to discern low-value from high-value practices.

To overcome these difficulties, we developed alternative approaches towards low-value and unnecessary care in the Netherlands. We surveyed Dutch GPs as we expected that they would be well-positioned to overview and assess the value of care throughout the system. It is not possible to directly verify these assessments due to the reasons above. However, we were actually surprised by the degree of consistency of the observed patterns; and such agreement/consensus strengthens our findings. In addition, we have developed a standardized approach to identify low-value services from medical practice guidelines; and as such, measurement of low-value care is only one step ahead.

Our second approach encompassed an exploration of the characteristics and utilization of high-cost patients. Our analysis was aimed at providing a patient-centric perspective towards costs, and to provide a comprehensive overview of high-cost patients' characteristics and utilization, in order to inform policy and intervention. The meaningfulness of such analyses improves when the breadth of service coverage increases, to fully understand drivers of high utilization across healthcare domains. One limitation of our Dutch research on high-cost patients is that our analyses were limited to the Health Insurance Act, as insurers argued that long term care data were of insufficient quality for our research purposes. Our research was partly aimed at overcoming this problem, and we focused our review towards studies that covered a broad range of services across the continuum of care at health system level, and excluded all studies with a narrow scope of costs and all studies with a narrow

population base. We prefer increasingly broad approaches in high-cost patients analyses above narrow approaches at local level. However, the latter analyses (see box 1, a high-cost patients analysis in one general hospital in the Netherlands) are also valuable on their own, and may actually address shortcomings of broader approaches, through their use of other data sources, and an increasing focus on subpopulations that were identified in the broad studies. Besides, local approaches may be more able to directly identify opportunities to intervene.

Broader context of our findings

The fiscal sustainability of healthcare depends on numerous factors. Chapter one and two provided an overview of (recent reforms in) the Dutch healthcare system. This thesis should be interpreted in the context of a range of other trends.

Aging and multimorbidity

The Dutch population is aging and as a result there are more elderly at a higher average age. It is known that increasing age is associated with a higher prevalence of common chronic conditions, and that the number of chronic diseases is nearly exponentially related to costs [5]. Our studies also show multimorbidity is the prime driver of high costs. Besides, our studies actually show that multimorbidity is ubiquitous in high-cost patients, and that multimorbidity is not merely limited to elderly. For example, one primary subgroup of high-cost patients are mental health high-cost patients, and these patients are known for their co-morbid somatic care needs and utilization. At present, medical practice guidelines in the Netherlands are too much focused on one single disease. In the English NHS, a multimorbidity guideline was developed with the aim of reducing treatment burden and unplanned care, and to improve quality of life by promoting shared decision making [6]. As such, the aim of this guideline is to stimulate person-centred care, or care that is based on what is important to each person in terms of treatments, health priorities, lifestyle and personal goals. Multimorbidity also comes with challenges for the organization of our health system; as multimorbid patients are very likely to get treatments from several practitioners across several healthcare domains, for increasingly complex medical needs. Consequently, high quality of care requires coordination and integration of care across health care domains; as well as coordination and integration within healthcare domains.

Evolving role of GPs in the Netherlands

Decades ago, GPs used to accompany their patients in hospitals, and used to be involved in in-hospital decision making; whilst anecdotal evidence tells that GPs nowadays are involved only to a minor extent in the care for chronic multimorbid (and high-cost) patients. In other words: there seems to be a gap between the GP practice and hospitals; and coordination and integration across healthcare domains is lacking. We have also found this in other research. In the 2017 International Health Policy Survey among Dutch elderly we found that coordination of care may be improved. Of all respondents who said that they needed help in the coordination of care, only 69% received help from the GP or other professional in the GP practice [7]. In a policy document in 2012, the professional association of GPs proposed to strengthen their role as coordinators and 'guides' in the health system [8], but the effects of

this proposal are unknown. In sum, in chapter three we have shown that GP's gatekeeping abilities may be limited, and above we have shown that their role in the treatment of complex patients might be strengthened.

Continuing innovation, concentration and specialisation

There is ever more innovation in the healthcare sector, and scientific evidence of the value of new innovations typically lags behind. There are thus ever more treatment alternatives available for which comparative benefits and costs may be largely unknown. Chapter five showed that many technologies enter the health system without formal assessment of the costs and benefits. For such innovations, there is a lack of evidence of the value of the treatments, and a more strict control of the entry of these innovations is warranted. Besides, the chapter showed that hospitals choose their particular key topics, procedures, or patients groups that they are willing to invest in; and that they disinvest in other services. There is a broad consensus that the *concentration and dispersion* of new technologies and expertise may increase quality of care, and may help guarantee the future financial sustainability of such innovations. However, experience in the past has shown concentration primarily serves organisational and professional interests, rather than quality of care [9]. For patients, accessibility to innovations is at stake, and from a financial perspective, increased concentration may result in higher prices for specialized services.

Parallel to this trend of concentration of specialised services is the continuing proliferation of scientific evidence, and the inability of professionals to keep up with the evidence base in the full breadth of their specialty. There is a trend towards 'super-specialization', which may come at costs of generic knowledge and competences. This may hold especially for patients with multimorbidity (and high costs) in an aging population. The Dutch Federacy of Medical Specialists has proposed to bend this trend (they aim to educate more 'generalists') [10], but the effects of this proposal remain to be seen.

Routine data collection and learning health systems

There is ever more data, and routinely collected data are used more and more to inform healthcare service provision and policy. In the United States, routinely collected data have been used to identify low-value services to stimulate the systematic reduction in use of services (deimplementation) [11]. Chapters seven and eighth are also examples of such work. In addition, in the Netherlands there have been analyses of medical practice variation, and clinical registries are used more and more to inform practice and policy. Taken together, however, progress clearly lags behind its potential use. In the Netherlands, there is a general lack in the interoperability of systems (each general practice, hospital, etc. has its own electronic health record system), and there are many legislative and operative hurdles for using the data.

In previous work, we have elaborated on two approaches of using routinely collected data to reduce unwarranted variation in the use of services [12]. Especially the 'Shapiro-method' may be applied to high-cost patients (see also below, and box 1). In this method, much emphasis is put on stimulating and convincing the professionals. Central to projects is a physician champion, a professional with high esteem who is being coached and supported by data-analysts and experts. Physicians are in the lead to adapt analyses, in order to discern

warranted from unwarranted variation, and to identify opportunities to intervene. In the context of high-cost patients, hospitals and specialists themselves may be able to segment and analyze their patient population, in order to reconfigure their service organisation to those in heaviest need. As such, this approach fits perfectly within the novel learning health systems paradigm, which means "A learning health system is designed to generate and apply the best evidence for the collaborative healthcare choices of each patient and provider; to drive the process of discovery as a natural outgrowth of patient care; and to ensure innovation, quality, safety, and value in healthcare" [13]. Routinely collected data are used to compensate for the drawbacks of the evidence based medicine paradigm which we have outlined above. The feasibility of such approaches, however, may be largely dependent upon local incentives, most notably a (financially) safe local environment.

Experience with high-cost patients elsewhere

In Canada and the United States many interventions have been taken to increase the value of care for high-need high-cost patients [14-23], and such initiatives might inform future initiatives in the Netherlands. Together, evaluations point out that there is no single approach that outperforms others, and that activities require adaptation to local contexts and populations. Besides, interventions heavily rely on data and IT systems, and follow integrated, holistic and patient-centred approaches across the continuum of care.

In Canada, Community Health Links were introduced to bring together healthcare providers to better coordinate care [24]. Health links are voluntary, self-organizing systems inspired by US ACOs, and multispecialty physician networks [25,26]. It was named a 'low rules' intervention and at its implementation, the Ministry sought to find a balance between structure and flexibility, to allow for change through improvisation. All health links provide an added coordinating service that aims to increase access and bring together patients' health and social care teams. It is said that participating patients all 1) have an individualized, coordinated care plan 2) have care providers who follow the plan 3) get support to take the right medications 4) are able to call a providers who knows them and is familiar with the personal situation.

In the US, ACOs appeared to reduce utilization and spending among high-cost patients [25]. In addition, Sherry et al examined five community-oriented programs that successfully improved care for high-need, high-cost patients. The five programs shared common attributes, including flexible financing, shared leadership, shared data, and a strong shared vision of commitment toward delivery of person-centered care. Other studies listed other sets of common attributes for successful programs, including closely targeting patients for intervention, comprehensive assessments of risks and needs, specially trained managers who facilitate coordination and communication and effective interdisciplinary teamwork [14,23]. One notable example is the Ambulatory Intensive Caring Unit; wherein high-cost patients receive all their care from a separate high-risk clinic or a high-risk team within a clinic. Patients no longer receive care from a primary care provider who sees both complex and non-complex patients, such that the entire attention of the team is focused on only a small number of high utilizing patients [27, 28]. Such an approach is now about to be taken in one hospital in the Netherlands (see box 1).

Box 1. Bernhoven and high-cost patients

Bernhoven reviewed the characteristics and healthcare utilization of their highest-cost patients. In their year of analysis, the top-1% of their population accounted for 20% of total costs. After identification of high-cost patients a sample of 55 patients was selected for systematic analysis. Experienced medical specialists reviewed patient health records, as comprehensive/integral as possible. A multidisciplinary team discussed the findings and discussed possible implications for practice.

Many patients were characterized by complex and advanced stages of disease, and analysis pointed to a small set of shared problems across the cases, that all have to do with a lack of coordination of care and lack of an integral view upon the patient's care needs. For some patients, the decision to proceed to a palliative trajectory posed problems; the geriatrician or palliative team were not involved, or too late. One problem was that 'agreed on policy' was not acted upon in practice. For example, patients were hospitalized against the advice of the palliative team and the patient's wish, or pre-terminal patients were admitted to the intensive care unit. Patients were often admitted to non-dominant specialisms (for example cardiologists in patients with heart failure, and other minor comorbidities), and the dominant specialists were sometimes unaware of the admission. Furthermore, patients received contra-indicated treatments (predominantly contra-indicated medications). In addition, the general practitioner was only to a limited extent involved in the treatment of the patient. He/she receives many letters from the hospital, but patients were not visiting to the GP anymore when they go to the hospital often.

Based on this information, Bernhoven is about to open a separate high-risk clinic for high-need high-cost patients (all patients following specific criteria). A generalist (either a internist-geriatrician, or geriatrician) and specialized nurse will run the clinic. Patients will no longer be followed routinely by remaining specialists, but the generalist is very much in control and is the one to request additional consultation if needed. The generalist will actively cooperate with the general practice, and palliative team.

Implications for policy and practice

Based on the broader context and our research findings, we drafted the following recommendations for policy and practice. Some recommendations are new for the Netherlands, and some build on ongoing initiatives that can be accelerated or extended. All recommendations should be interpreted as broad directions for policy that need further validation.

Societal level:

- To support integrated care for complex patients, data systems need to be improved, so that practitioners can get a complete insight into the healthcare use and medical records of patients. In the short term this requires interoperability of regional health records; in the longer term a national health record or personal health environment could

provide for this. The facilities must also be able to communicate a patient's care plan, and the patient's perspective on his or her life. Such data systems may also facilitate the identification of low-value services.

- It is important that much more attention is paid to regional coordination and cooperation between the different levels (vertical networks), such as university medical centres, top clinical hospitals, general hospitals, general practices, allied healthcare, and the social domain. This also requires guidance from health insurers who can play a mediating role here.
- At the national level, more attention is warranted to the theme of multimorbidity. To support general practitioners (who are too busy, and may have insufficient knowledge and facilities) and geriatricians (specialists of the elderly and geriatric syndromes, not multimorbidity in particular), more generalists need to be trained, such as 'hospital doctors' or 'multimorbidity doctors' [29]. In addition, more attention should be paid to multimorbidity in new (or updates of) medical practice guidelines. In addition, a separate multimorbidity guideline could be developed for hospital specialists, in accordance with the guideline in the English NHS. Based on this guideline, doctors are able to not comply with disease-specific guideline recommendations, if necessary.
- The 'open' benefit package for non-pharmaceutical innovations could be more 'closed', to prevent widespread use of questionable services that lack a solid evidence base, such as Da Vinci surgery. Stakeholders could cooperate more, to 'guide' the introduction of new innovations. For example, minimum quality requirements could be established. Coverage with evidence development could also be used for non-pharmaceuticals. Although health insurers increasingly pay attention to the entry of innovations, a more active purchasing policy seems justified.

Local/regional level:

- Our do-not-do list has been integrated in the website that presents all medical practice guidelines to medical specialists. In addition, in the development of new medical practice guidelines attention is being paid to define new do-not-do recommendations, in order to stimulate disinvestment of such activities. To further stimulate deimplementation, the value of treatments could be discussed much more, and much more critically at local levels. Insurers might opt to stop funding low-value services.
- Patient selection for major procedures or expensive medicines can be improved, not only in the elderly. There are several options for this, including geriatric screening, or multidisciplinary decision-making which helps to prevent seeking 'the edges of the indication criteria' or that the treatment choice does not match the preferences of the patient.
- Individual providers, such as general practitioners, hospitals (or their departments), may analyze and segment their top 1% or top 5% patient population themselves, to inform policy and practice. An analysis based on administrative claims may be sufficient for this. However, such analyses can also be performed bottom-up, by implementing the *5x5x5 method*. A provider identifies five random patients with particularly high healthcare costs (costs are included as widely as possible) and analyzes as comprehensive as possible (in an integrated way, from multiple angles, all treating professionals, the patient, informal

carer) and as accurately as possible what the care process of these patients looked like, what went well and what could have gone better. On the basis of the first five cases, shared problems are identified and improvement measures can be taken. Later on, the project can be repeated but for 25 patients and later for 125 patients. Box one shows how Bernhoven has applied this method and what steps they took on basis of their findings. This is a perfect solution in a search for horizontal integration, alignment of care within one organisation or organisational level. Patients with multimorbidity are diverse populations, which requires flexibility in the organisation of their services to organize the care according to their particular needs.

- More attention should be paid to high quality transitional care for hospitalized patients. This is currently lacking, partly because the responsibility for this is not clearly stated. Activities such as the 'Transmurale zorgbrug' could be scaled up, and may also be extended beyond frail elderly. GPs primarily play a role stepped-down for these patients, for organizing and providing good post-discharge care. In addition, GPs may find their way back into the hospital, to accompany their patients as authoritative advisor in complicated treatment decisions, in order to stimulate treatment decisions that best fit with the patient's preferences and social context.
- GPs and medical specialists might receive additional training for the treatment of complex patients. Training might be developed according to the 'Ariadne' principles , 'collaborative goal setting', and shared decision making [30-33]. In the care of vulnerable patients, multiple professionals are involved and irrevocably, problems arise with respect to the mutual division of responsibilities. It is important that such issues are acknowledged and that professionals agree on a set of processes about how to deal with the patients. This includes informing each other much more, and more (multidisciplinary) discussion about the appropriateness of alternative treatment options.

Implications for future research

This thesis has provided a solid base for deimplementation projects, and the evaluation of these projects might powerfully inform larger projects at the national level. Further research is needed to identify low-value services in practice, preferably on the basis of routinely collected data. In addition, as the identification of low-value care is only one of several necessary steps, more research is needed on how to effectively reduce low-value services. Future approaches might consider to combine current approaches with financial incentives.

Above we suggested that patient selection for major procedures or expensive medicines might be improved through geriatric screening, or multidisciplinary decision-making. One notable example of this is the AGE-CRC-study, that aims to develop a pre-operative prediction-model in order to prevent under- and overtreatment in colon cancer [34]. Similarly, a multidisciplinary approach proved to reduce inappropriate transaortic valve replacements [35]. Alternatives such as patient selection by a professional that is not the surgeon may conflict with the professional autonomy of doctors. Further research is needed to determine feasibility of such measures, and if such measures can contribute to keeping healthcare affordable and reduce unnecessary treatments.

We have shown that multimorbidity is a prime driver of high costs, that multimorbid patients are likely to incur high hospitals costs, and that these patients may benefit from

person-centred and integrated healthcare. Future studies might investigate whether integrated care models within the hospital, or with the hospital as a locus (box 1 shows one example of an integrated care initiative with the hospital as a locus), may contribute to keeping healthcare affordable. Above we argued that there is a need for more vertical as well as horizontal integration. Further research is needed to investigate how such networks are best developed and what (contextual) factors stimulate or discourage the process of efficient network development.

We have also shown that care improvement programs need adaptation to local contexts and populations, and that such programs follow integrated approaches across the continuum of care. Data and IT systems can be used to identify target populations, to align providers and provide them with reference data, and to inform the continuous development of the program. Zulman et al recently published about partnered research in healthcare delivery redesign for high-cost patients. In this approach researchers firstly analyze healthcare use and characteristics of high-cost patients, and perform stakeholder need assessments to inform the redesign of healthcare delivery [36]. Canadian work showed that complex adaptive systems (CAS) theory may have strong potential to understand and support policy design and implementation. The theory views healthcare as numerous subsystems characterized by diverse agents that interact, self-organize, and continuously adapt; and is used to describe systems that cannot be understood in their entirety as a result of many interacting variables and forces. As such, it is useful for understanding the implementation of integrated networks. According to complex adaptive systems theory, initiatives should enhance scope for new interconnections, sensemaking, self-organization, emergence, and co-evolution [24]. More generally, in learning health systems, patient segmentation analysis about the characteristics in healthcare utilization of high-cost patients may be pivotal [37], and there is a need for interdisciplinary work using a variety of both quantitative and qualitative research. Such qualitative work is needed to provide quantitative analysis with the right context and processes, to align stakeholders, and to further inform future quantitative work.

Conclusion

In this thesis we explored two approaches for reducing unnecessary and low-value care while simultaneously improving outcomes of care, and reducing spending. An interdisciplinary approach was taken through a variety of research methods, including survey research, document analysis, a qualitative interview study, a systematic review, and claim database analyses. Our results show that that there is ample room for quality improvement and cost reduction. For low-value services, continuing reassessment and discussions are needed to identify those services that may be of little value, and to inform concomitant policy and intervention, in order to reduce unnecessary spending. A myriad of policy alternatives are available for redesigning our health system according to the needs of the patients in heaviest need for high-quality healthcare, and to reduce unnecessary spending. Both approaches are best informed through multidisciplinary research, that include both quantitative as well as qualitative work and engagement of professionals to inform local redesign.

References

1 Landelijke Huisartsenvereniging (LHV), InEen (vereniging van organisaties voor eerstelijns zorg), Patiëntenfederatie Nederland, Zorgverzekeraars Nederland (ZN), De minister voor Medische Zorg en Sport (VWS). Onderhandelaarsakkoord huisartsenzorg 2019 t/m 2022. 2018.
2 Garrow JS. What to do about CAM: How much of orthodox medicine is evidence based? BMJ. 2007;335(7627):951.
3 Figueroa JF, Joynt Maddox KE, Beaulieu N, Wild RC, Jha AK. Concentration of Potentially Preventable Spending Among High-Cost Medicare Subpopulations: An Observational Study. Annals of internal medicine. 2017.
4 McWilliams JM, Schwartz AL. Focusing on High-Cost Patients – The Key to Addressing High Costs? The New England journal of medicine. 2017;376(9):807-9.
5 Lehnert T, Heider D, Leicht H, Heinrich S, Corrieri S, Luppa M, et al. Review: health care utilization and costs of elderly persons with multiple chronic conditions. Medical care research and review : MCRR. 2011;68(4):387-420.
6 NICE. Multimorbidity: clinical assessment and management. 2016.
7 Wammes JJG, van der Wees P, Westert GP. International Health Policy Survey 2017. Onderzoek onder 65-plussers in 11 landen. Nijmegen: Scientific Institute for Quality in Healthcare (IQ healthcare), Radboudumc; 2017.
8 NHG, LHV. Toekomstvisie huisartsenzorg 2022. 2012.
9 Zuiderent-Jerak T, Kool T, Rademakers J. De relatie tussen volume en kwaliteit van zorg: tijd voor een brede benadering. Utrecht/Nijmegen/Rotterdam.: Consortium onderzoek kwaliteit van zorg; 2012.
10 Federatie medisch specialisten. Visiedocument Medisch Specialist 2025 Utrecht; 2017.
11 Schwartz AL, Chernew ME, Landon BE, McWilliams JM. Changes in Low-Value Services in Year 1 of the Medicare Pioneer Accountable Care Organization Program. JAMA Intern Med. 2015;175(11):1815-25.
12 Wammes JJG, Atsma F, de Man Y, Groenewoud S, Westert GP. De zorg is niet overal hetzelfde; het reduceren van ongewenste verschillen in kwaliteit en kosten in Nederland?. Betaalbare zorg. Nijmegen: Celsus academie voor betaalbare zorg; 2018.
13 Institute of Medicine (US). Roundtable on Evidence-Based Medicine, the Learning Healthcare System: Workshop Summary. Washington (DC): National Academy Press (US); 2007.
14 Anderson GF, Ballreich J, Bleich S, Boyd C, DuGoff E, Leff B, et al. Attributes common to programs that successfully treat high-need, high-cost individuals. American Journal of Managed Care. 2015;21(11):e597-e600.
15 Blumenthal D, Abrams MK. Tailoring Complex Care Management for High-Need, High-Cost Patients. Jama. 2016.
16 Blumenthal D, Chernof B, Fulmer T, Lumpkin J, Selberg J. Caring for High-Need, High-Cost Patients – An Urgent Priority. The New England journal of medicine. 2016;375(10):909-11.
17 Brown RS, Peikes D, Peterson G, Schore J, Razafindrakoto CM. Six features of Medicare coordinated care demonstration programs that cut hospital admissions of high-risk patients. Health affairs. 2012;31(6):1156-66.

18 Gawande A. The hot spotters: can we lower medical costs by giving the neediest patients better care? The New Yorker. 2011.
19 Hong CS, Siegel AL, Ferris TG. Caring for high-need, high-cost patients: what makes for a successful care management program? Issue brief. 2014;19:1-19.
20 Sherry M, Wolff JL, Ballreich J, DuGoff E, Davis K, Anderson G. Bridging the Silos of Service Delivery for High-Need, High-Cost Individuals. Population health management. 2016.
21 Vrijhoef B, Thorlby R. Developing care for a changing population: supporting patients with costly, complex needs. Nuffield Trust; 2016.
22 Bleich SN, Sherrod C, Chiang A, Boyd C, Wolff J, DuGoff E, et al. Systematic Review of Programs Treating High-Need and High-Cost People With Multiple Chronic Diseases or Disabilities in the United States, 2008-2014. Prev Chronic Dis. 2015;12:E197.
23 Blumenthal D, Anderson G, Burke SP, Fulmer T, Jha AK, Long P. Tailoring Complex-Care Management, Coordination, and Integration for High-Need, High-Cost Patients, 2016.; 2016.
24 Grudniewicz A, Tenbensel T, Evans JM, Steele Gray C, Baker GR, Wodchis WP. 'Complexity-compatible' policy for integrated care? Lessons from the implementation of Ontario's Health Links. Soc Sci Med. 2018;198:95-102.
25 Hsu J, Price M, Vogeli C, Brand R, Chernew ME, Chaguturu SK, et al. Bending The Spending Curve By Altering Care Delivery Patterns: The Role Of Care Management Within A Pioneer ACO. Health affairs. 2017;36(5):876-84.
26 Stukel TA, Glazier RH, Schultz SE, Guan J, Zagorski BM, Gozdyra P, et al. Multispecialty physician networks in Ontario. Open Med. 2013;7(2):e40-55.
27 Milstein A, Gilbertson E. American medical home runs. Health affairs. 2009;28(5):1317-26.
28 Bodenheimer T. Strategies to Reduce Costs and Improve Care for High-Utilizing Medicaid Patients: Reflections on Pioneering Programs. Centre for Health Care Strategies; 2013.
29 Plochg T, Keijsers JF, Levi MM. [The 'multimorbidity generalist' is the future]. Ned Tijdschr Geneeskd. 2012;156(42):A5515.
30 Vermunt N, Harmsen M, Westert GP, Olde Rikkert MGM, Faber MJ. Collaborative goal setting with elderly patients with chronic disease or multimorbidity: a systematic review. BMC Geriatr. 2017;17(1):167.
31 Bower P. Better management of multimorbidity: a critical look at the 'Ariadne principles'. BMC Med. 2014;12:222.
32 Muth C, van den Akker M, Blom JW, Mallen CD, Rochon J, Schellevis FG, et al. The Ariadne principles: how to handle multimorbidity in primary care consultations. BMC Med. 2014;12:223.
33 Prados-Torres A, Del Cura-Gonzalez I, Prados-Torres JD, Leiva-Fernandez F, Lopez-Rodriguez JA, Calderon-Larranaga A, et al. [Multimorbidity in general practice and the Ariadne principles. A person-centred approach]. Aten Primaria. 2017;49(5):300-7.
34 IKNL, Prospectief Landelijk CRC cohort (colon cancer). AGE-CRC-studie: betere kwaliteit van leven voor ouderen met darmkanker 2017 [Available from: https://www.iknl.nl/over-iknl/nieuws/nieuws-detail/2017/11/13/age-crc-studie-betere-kwaliteit-van-leven-voor-ouderen-met-darmkanker.

35 Schoon Y, de Boer M, Olde Rikkert M. Niet elke oudere heeft baat bij nieuwe hartklep. 2015.
36 Zulman DM, Ezeji-Okoye SC, Shaw JG, Hummel DL, Holloway KS, Smither SF, et al. Partnered research in healthcare delivery redesign for high-need, high-cost patients: development and feasibility of an Intensive Management Patient-Aligned Care Team (ImPACT). J Gen Intern Med. 2014;29 Suppl 4:861-9.
37 Vuik SI, Mayer EK, Darzi A. Patient Segmentation Analysis Offers Significant Benefits For Integrated Care And Support. Health affairs. 2016;35(5):769-75.

Summary

The studies in this thesis explored two approaches for reducing unnecessary and possibly harmful care – so-called low-value services – while simultaneously improving outcomes of care, and reducing spending. As such, this thesis was aimed at improving the fiscal sustainability of healthcare. Our research questions were:

1 What are opportunities for cost-reduction through reduction of low-value services in the Netherlands?
2 What are the characteristics and healthcare utilization of high-cost patients and what strategies do likely improve high-cost patients' care and reduce costs?

In line with the research questions, this thesis is divided into two parts. The first part about low-value services and unnecessary care is covered in chapters 3-5. The second part about high-cost patients is covered in chapters 6-8. Chapters 1 and 2 provide necessary introductory information, but will not be discussed below. Below we summarize the research results of this thesis and its implications for policy and practice, and research.

Chapter 3 presents an exploratory survey among Dutch GPs. We found that, according to Dutch GPs, patients receive too much care in general hospitals, in primary care, in GP cooperatives as well as in private clinics. The Dutch GPs' demand-satisfying attitude and the increased availability of diagnostic facilities most saliently contribute to the provision of excess care at the entry point of care in the Netherlands. Also misaligned incentives induce that Dutch GPs may not sufficiently pick up the gatekeeping role. Our results show practitioners often find it difficult to deny enduring patients access to further care, even if they think treatment is unnecessary from a medical point of view. This creates an image of GPs acting in a demand-satisfying way in their referrals and treatment decisions. Besides, our results indicate that GPs themselves are prepared to avoid unnecessary hospital care - versus reducing unnecessary care in primary care - yet that the preconditions at the level of the health system do not meet. For example, GPs found that funding gaps between primary care and hospitals impede cooperation and coordination, and that this provokes unnecessary care. This chapter concludes that discussion and exploration by GPs and policy makers about the complicated and sometimes unintended effects of strengthening primary care and its interactions with unnecessary care may be fruitful.

Chapter 4 describes the development of a list of lower value services identified from 193 Dutch clinical practice guidelines, published between 2010 and 2015. In total, 1366 lower value services were extracted from 193 Dutch guidelines. Of the lower value services 30% covered diagnostics, 29% related to non-drug treatment and 39% to drug treatment. The majority (77%) of all low-value services was on care that should not be offered at all, whereas the other 23% recommended on care that should not be offered routinely. ICD10-chapters that included most lower value services were neoplasms and diseases of the nervous system. This chapter concluded that the development of a comprehensive list of lower value services and prioritization is only the first of several necessary steps in reducing low-value services.

Chapter 5 shows the results of our interview study of displacement effects in Dutch hospitals. We studied how cost-increasing services have entered Dutch hospitals and what services were displaced to accommodate the entry of these services. Our findings show that it is difficult to identify the services that are displaced to accommodate the cost-increasing health technologies; limited transparency in the flow of funds within a hospital contributed to this. Besides, we found that the entry of new innovations and cost-containment are two parallel processes that are generally not causally linked. The way of financing is pivotal in displacement in the Netherlands. The budget pressure of expensive drugs (that amounts to a separate budget, not part of department budgets) is linked to horizontal reallocation across departments, whereas the budget pressure of remaining services is linked to vertical reallocation within departments or divisions. This chapter concludes that hospitals ration mainly in response to cumulative cost pressures, production ceilings and capacity problems, and that active surveillance of waiting lists is warranted to prevent waiting list driven morbidity.

Chapter 6 presents the results of our systematic review of high-cost patients' characteristics and healthcare utilization. We used Andersen's behavioral model to organize the findings. Our results indicate that across health systems and nations, a high prevalence of multiple (chronic) conditions consistently explain high-cost patients' utilization. Besides, we found a high prevalence of mental illness across all the studies, most notably in US Medicaid and total population studies. We found that various health system characteristics may contribute to high costs, and that preventable spending was estimated at maximally ten percent of spending. Furthermore, we found that high costs are associated with increasing age and that clinical diagnoses and utilization patterns varied across age groups. However, still more than half of high-cost patients are younger than 65 years. High costs were associated with higher incomes in the US, but with lower incomes elsewhere. Finally, we confirmed that high-cost patients are more likely to die, and decedents are more likely to incur high-costs. However, no more than 30% of high-cost patients were in their last year of life. This chapter concluded that high-cost patients make up the sickest and most complex populations and that their high utilization is primarily explained by high levels of chronic and mental illness.

Chapter 7 presents our Dutch claim database study of high-cost patients' characteristics and healthcare utilization. We found that expensive treatments, most cost-incurring condition, and age proved to be informative variables for studying this heterogeneous population. Expensive care use (expensive drugs, ICU treatment, dialysis, transplant care, DRG > €30,000) contributed to high costs in one third of top-1% beneficiaries and in less than 10% of top-2-5% beneficiaries. High-cost beneficiaries were overwhelmingly treated for diseases of circulatory system, neoplasms, and mental disorders. More than 50% of high-cost beneficiaries were 65 years of age or younger, and average costs decreased sharply with higher age within the top-1% population. This chapter concludes that high-cost patients are usually treated for several conditions and use care from multiple providers, and that tailored interventions are needed to meet the needs of high-cost beneficiaries, and to avoid waste of scarce resources.

Chapter 8 presents our claim database study of patients with heart failure and high costs. We found that more than 90% of patients with heart failure incurred at least one top 5% year, and 32% incurred at least one top 1% year. Besides, top-1% utilization predominantly occurs incidentally, whereas more than half experience two or more top 5% years and the majority of these top 5% years were incurred consecutively. Patients with heart failure and top 1% and top 2-5% utilization differed from others in their higher rate of chronic conditions and multimorbidity, excessive polypharmacy, hospital admissions, and heart-related surgeries. In addition, the top 1% group experienced a remarkable increase of mental health care costs during the initial year with heart failure. This chapter concludes that comprehensive and integrated efforts are needed to further improve quality of care and reduce unnecessary costs.

Following the results in this thesis, we drafted a range of recommendations for policy and practice. Below we present a selection. First of all, to support integrated care for complex patients, data systems need to be improved, so that practitioners can get a complete insight into the healthcare use and medical records of patients. Such data systems may also facilitate the identification of low-value services. At the national level, more attention is warranted to the theme of multimorbidity. We argue that there is a need for more generalists, and more attention to multimorbidity in medical practice guidelines. Furthermore, we suggest the 'open' benefit package for non-pharmaceutical innovations could be more 'closed', for example through coverage with evidence development for non-pharmaceuticals. Furthermore, we argue for more (multidisciplinary) discussion about the value of treatments, to stimulate the deimplementation of low-value services. In addition, we suggest that GPs may find their way back into the hospital, to accompany their patients as authoritative advisor in complicated treatment decisions, in order to stimulate treatment decisions that best fit with the patient's preferences and social context.

Samenvatting

Patiënten met hoge zorgkosten en mogelijkheden om onnodige uitgaven te verminderen

Dit proefschrift had als doel te onderzoeken hoe onnodige en mogelijk schadelijke zorg vast te stellen, om het gebruik hiervan te verminderen en de uitgaven te verminderen. Als zodanig was dit proefschrift gericht op het verbeteren van de betaalbaarheid van de gezondheidszorg. De onderzoeksvragen waren:

1 Wat zijn mogelijkheden voor kostenbeheersing door vermindering van onnodige zorg in Nederland?
2 Wat zijn de karakteristieken en het zorggebruik van de patiënten met hoogste zorgkosten en welke strategieën verbeteren de zorg voor deze patiënten en verlagen de kosten?

Dit proefschrift is verdeeld in twee delen. Het eerste deel heeft betrekking op onnodige zorg en wordt behandeld in hoofdstukken 3 tot en met 5. Het tweede deel gaat over patiënten met hoge zorgkosten en wordt behandeld in hoofdstukken 6 tot en met 8. Hoofdstukken 1 en 2 bieden noodzakelijke inleidende informatie, maar zullen hieronder niet worden besproken. Hieronder vatten we de resultaten van dit proefschrift samen en bespreken we de implicaties voor onderzoek, beleid en praktijk.

Hoofdstuk 3 presenteert een survey-onderzoek onder Nederlandse huisartsen. Nederlandse huisartsen vonden dat patiënten te veel zorg ontvangen in ziekenhuizen, in de eerstelijn, in huisartsenposten en in privéklinieken. De vraaggerichte houding van Nederlandse huisartsen en de toegenomen beschikbaarheid van diagnostische faciliteiten dragen bij aan de verlening van onnodige zorg in de eerstelijn in Nederland. Huisartsen vinden het vaak moeilijk om vasthoudende patiënten de toegang tot verdere zorg te ontzeggen, zelfs als zij van mening zijn dat dit vanuit medisch oogpunt niet nodig is. Dit creëert een beeld van huisartsen die op een vraaggerichte manier handelen bij hun verwijzingen en behandelbeslissingen. Onze resultaten duiden er ook op dat huisartsen bereid zijn onnodige ziekenhuiszorg te voorkomen, maar dat zij hierin ook gehinderd worden door ontbrekende randvoorwaarden op het niveau van het gezondheidssysteem. Huisartsen gaven aan dat financieringsschotten een belemmering vormen voor samenwerking en coördinatie tussen de eerstelijnszorg en ziekenhuizen, met onnodige zorg als gevolg. Er is behoefte aan meer onderzoek naar, en discussie over de gecompliceerde en soms onbedoelde effecten van versterking van de eerstelijn en de interacties die dit heeft met onnodige zorg.

Hoofdstuk 4 beschrijft de ontwikkeling van de Beter-niet-doen-lijst, een lijst van zorghandelingen met weinig toegevoegde waarde geïdentificeerd uit 193 Nederlandse klinische richtlijnen, die waren gepubliceerd tussen 2010 en 2015. In totaal werden 1366 handelingen met weinig toegevoegde waarde geïdentificeerd. Van deze handelingen betrof 30% diagnostiek, 29% had betrekking op niet-medicamenteuze behandeling en 39% op medicamenteuze behandeling. De meerderheid (77%) van alle handelingen met weinig

toegevoegde waarde had betrekking op zorg die helemaal niet aangeboden zou moeten worden, terwijl de andere 23% zorg betrof die niet-routinematig moet worden aangeboden. ICD10-hoofdstukken met de meeste handelingen met weinig toegevoegde waarde waren nieuwvormingen en ziekten van het zenuwstelsel. De ontwikkeling van een lijst van handelingen met weinig toegevoegde waarde en prioritering hiervan is slechts een eerste van verschillende noodzakelijke stappen om deze handelingen te verminderen.

Hoofdstuk 5 toont de resultaten van ons interviewonderzoek naar verdringingseffecten in Nederlandse ziekenhuizen. We hebben onderzocht hoe kostenverhogende innovaties hun intrede deden in Nederlandse ziekenhuizen en welke zorg is 'verdrongen' om de intrede van de innovatie mogelijk te maken. Onze bevindingen tonen aan dat het moeilijk is specifieke zorghandelingen aan te wijzen die zijn verdrongen om ruimte te bieden aan kostenverhogende innovaties; beperkte transparantie in de geldstromen binnen ziekenhuizen heeft hieraan bijgedragen. Bovendien ontdekten we dat de intrede van nieuwe innovaties en kostenbeheersing twee parallelle processen zijn die over het algemeen niet met elkaar zijn verbonden. Wij vonden dat de manier van financiering cruciaal is bij verdringing in Nederland. De kostendruk van dure geneesmiddelen (dat een afzonderlijk budget vormt, geen deel van afdelingsbudgetten) is gerelateerd aan horizontale herallocatie tussen afdelingen, terwijl de budgetdruk van de resterende zorg is gerelateerd aan verticale herallocatie binnen afdelingen of divisies. Ziekenhuizen rantsoeneren voornamelijk ten gevolge van cumulatieve budgetdruk, productieplafonds en capaciteitsproblemen. Actieve monitoring van wachtlijsten is gerechtvaardigd om nadelige effecten ten gevolge van wachtlijsten zoveel mogelijk te voorkomen.

Hoofdstuk 6 presenteert de resultaten van ons systematisch literatuuronderzoek naar de karakteristieken en zorggebruik van patiënten met hoge zorgkosten. Wij maakten hierin een vergelijking tussen verschillende landen en zorgsystemen. We gebruikten het behavioral model van Ronald Andersen om de bevindingen te ordenen. Onze resultaten laten zien dat in alle onderzochte zorgstelsels en landen een hoge prevalentie van meerdere (chronische) aandoeningen consistent het gebruik van hoge kosten verklaarde. Bovendien vonden we een hoge prevalentie van psychische aandoeningen. We ontdekten dat verschillende kenmerken van het gezondheidssysteem kunnen bijdragen aan hoge kosten. Maximaal tien procent van de uitgaven waren 'vermijdbaar'. Bovendien hebben we geconstateerd dat hoge zorgkosten geassocieerd zijn met toenemende leeftijd en dat de kenmerkende klinische diagnoses en patronen in zorggebruik variëren per leeftijdsgroep. Echter, nog steeds is meer dan de helft van patiënten met hoge zorgkosten jonger dan 65 jaar. Hoge kosten waren geassocieerd met hogere inkomens in de VS, maar juist met lagere inkomens in overige landen. Tenslotte vonden we dat patiënten met hoge zorgkosten meer kans lopen om te overlijden, en dat overledenen vaker hoge kosten hebben. Echter, niet meer dan 30% van de patiënten met hoge zorgkosten was in hun laatste levensjaar. Patiënten met hoge zorgkosten zijn de ziekste en meest complexe populaties. Hun hoge zorggebruik wordt voornamelijk verklaard door een hoog niveau van chronische en psychische aandoeningen.

Hoofdstuk 7 presenteert ons Nederlandse longitudinale databaseonderzoek naar de karakteristieken en zorggebruik van patiënten met hoge zorgkosten. We vonden dat dure behandelingen, de meest kostbare aandoening en leeftijd informatieve variabelen zijn voor het bestuderen van deze heterogene populatie. Dure zorgvoorzieningen (dure geneesmiddelen, ICU-behandeling, dialyse, transplantatiezorg, DBC > € 30.000) droegen bij aan hoge kosten bij een derde van de top 1% patiënten en bij minder dan 10% van de top 2-5% patiënten. Patiënten met hoge zorgkosten werden voornamelijk behandeld voor cardiovasculaire aandoeningen, nieuwvormingen en psychische- en gedragsstoornissen. Meer dan 50% van de patiënten met hoge zorgkosten was 65 jaar of jonger en de gemiddelde kosten daalden scherp met een hogere leeftijd binnen de top 1% groep. Patiënten met hoge zorgkosten worden vaak behandeld voor meerdere aandoeningen en gebruiken zorg bij meerdere aanbieders. Gerichte interventies zijn nodig voor het verbeteren van de zorg aan patiënten met hoge zorgkosten en om verspilling van schaarse middelen te voorkomen.

Hoofdstuk 8 presenteert onze databasestudie naar patiënten met hartfalen en hoge zorgkosten. We ontdekten dat meer dan 90% van de patiënten met hartfalen minstens één top 5% jaar had en dat 32% minstens één top 1% jaar had. Bovendien vonden wij dat top 1% gebruik overwegend incidenteel plaatsvindt, terwijl meer dan de helft van de patiënten met hartfalen meerdere top 5% jaren ervaart. Het merendeel van deze top 5% jaren vindt achtereenvolgens plaats. Patiënten met hartfalen en hoge zorgkosten verschilden van andere patiënten met hartfalen op tal van kenmerken, waaronder het aantal chronische aandoeningen, multimorbiditeit en het percentage met overmatige polyfarmacie, ziekenhuisopnames en hartgerelateerde operaties. Bovendien kende de top 1% groep een opmerkelijke stijging van de kosten voor geestelijke gezondheidszorg. Er zijn geïntegreerde inspanningen nodig om de kwaliteit van zorg verder te verbeteren en onnodige kosten te verminderen.

Op basis van de bevindingen van dit proefschrift hebben wij een reeks aanbevelingen opgesteld voor beleid en praktijk. Hieronder presenteren we de belangrijkste aanbevelingen. Verbeterde informatievoorziening binnen en tussen zorgaanbieders is nodig om goede geïntegreerde zorg voor complexe patiënten te ondersteunen, zodat behandelaars een volledig inzicht kunnen krijgen in de medische dossiers en het zorggebruik van hun patiënten. Dergelijke informatievoorzieningen kunnen ook de identificatie van zorg met weinig toegevoegde waarde vergemakkelijken. Op nationaal niveau is meer aandacht nodig voor het thema multimorbiditeit. Er behoefte aan meer generalisten en meer aandacht voor multimorbiditeit in medische richtlijnen. Verder stellen we voor dat het ‘open’ pakket voor niet-farmaceutische medische zorg meer ‘gesloten’ zou kunnen worden, bijvoorbeeld door sluisconstructies te ontwikkelen voor kostbare niet-farmaceutische producten en interventies. Verder pleiten we voor meer (multidisciplinaire) discussies over de waarde van behandelingen. Tot slot stellen we voor dat huisartsen meer intensief contact hebben met het ziekenhuis om hun patiënten te begeleiden als gezaghebbend adviseur bij ingewikkelde behandelbeslissingen.

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Curriculum Vitae

Joost Wammes was born on the 1st of January 1987 in Beusichem, the Netherlands. He grew up in Beusichem and in 2005 he completed his VWO (Preparatory Scientific Education) at the Koningin Wilhelmina College in Culemborg. He subsequently studied Biomedical Sciences at the Radboud University in Nijmegen and obtained his Master degree in 2011. He specialized in Health Technology Assessment and took a minor in business and economics at the University of Utrecht.

In late 2011, he started working as a researcher at IQ healthcare (Radboudumc) and was involved in numerous projects related to quality in healthcare. In 2013, IQ healthcare and the Ministry of Health founded the Celsus academy for sustainable healthcare, and Joost was involved in the first projects. In 2014, Joost started his PhD as part of the Celsus program, of which the results are described in this PhD thesis.

In 2018, he took a position at Logex, a healthcare analytics company that primarily works for and with Dutch hospitals and medical specialists.

Bibliography

Dutch

E Adang, N Stadhouders, C Parsons, **JJG Wammes**, P Govaert, D Determann, X Koolman, A Paulus, S Evers, G Frederix, W Oortwijn. Verdringingseffecten binnen het Nederlandse zorgstelsel. Radboudumc. 2018.

JJG Wammes. H18 Patiënten met intensieve zorgbehoeften. In: Betaalbare zorg. Jeurissen et al.

JJG Wammes, F Atsma, Y de Man, S Groenewoud, G Westert. H17 De zorg is niet overal hetzelfde; het reduceren van ongewenste verschillen in kwaliteit en kosten in Nederland In: Betaalbare zorg. Jeurissen et al.

N Stadhouders, **JJG Wammes**, E Adang. H15 'Verdringingseffecten' in de ziekenhuiszorg. In: Betaalbare zorg. Jeurissen et al.

JJG Wammes, P van der Wees, G Westert. International Health Policy Survey 2017. Commonwealth Fund. Onderzoek onder 65-plussers in 11 landen. IQ Healthcare 2017.

JJG Wammes, M Tanke, W Jonkers, G Westert, P van der Wees, P Jeurissen. Kenmerken en zorggebruik van patiënten met de hoogste zorgkosten. Nederlands Tijdschrift voor Geneeskunde. 2017.

JJG Wammes, N Stadhouders, W van Dijk, P Jeurissen. Ambities waargemaakt? Tien jaar Zorgverzekeringswet. Kwaliteit in zorg. 2017.

JJG Wammes, P van der Wees, J Burgers, P van Assendelft. Nederlandse arts druk met ggz, palliatie én bureaucratie. Medisch contact. 2016.

JJG Wammes, S Groenewoud, F Atsma, G Westert. Handleiding onderzoek naar praktijkvariatie in de gezondheidszorg. IQ healthcare. 2016.

JJG Wammes, E van den Akker-van Marle, T van Asselt, E Verkerk, S van Dulmen, T Kool. De Beter-niet-doen-lijst. Welke zorg dient niet(-routinematig) aangeboden te worden? IQ healthcare. 2016.

JJG Wammes, P van der Wees, G Westert. International Health Policy Survey 2016. Commonwealth Fund. Onderzoek onder volwassenen in 11 landen. IQ Healthcare 2016.

G Westert, P Jeurissen, M van den Berg, met mede werking van **JJG Wammes**. Hoofdstuk 10 De Nederlandse gezondheidszorg: werking en prestaties. Volksgezondheid en gezondheidszorg.

Team zorgatlas, waaronder **JJG Wammes**. Infographic rughernia etc. Bijvoorbeeld: http://www.sin-nl.org/wp-content/uploads/2014/01/NPCF-infographic-Rughernia-27-1-2014-20140122.pdf. IQ Healthcare, 2014, 2015.

JJG Wammes, P van der Wees, G Westert. International Health Policy Survey 2015. Commonwealth Fund. Onderzoek onder huisartsen in 11 landen. IQ Healthcare 2015.

JJG Wammes, R Akkermans, J Koetsenruijter, G Hannink, G Westert, P van der Wees. PROMs orthopedie – primaire knieoperaties. Op weg naar transparantie. IQ healthcare 2014.

JJG Wammes, R Akkermans, J Koetsenruijter, G Hannink, G Westert, P van der Wees. PROMs orthopedie – primaire en revisie heupoperaties. Op weg naar transparantie. IQ healthcare 2014.

J Wilschut, **JJG Wammes**, P Jeurissen. Kostenbeheersing dure zorgvoorzieningen. Internationale vergelijking van beheersinstrumenten bij de introductie van dure zorgvoorzieningen. Celsus academie voor betaalbare zorg. 2014.

JJG Wammes, S Groenewoud, W Woertman, E Adang. Budget Impact Analyse bij de update van de richtlijn Dementie. IQ Healthcare. 2014.

P van der Wees, **JJG Wammes**, G Westert, P Jeurissen. Omvang en financiering van het basispakket. Een verkennende internationale vergelijking. Celsus academie voor betaalbare zorg. 2013.

JJG Wammes, L Verhoef, G Westert, P Assendelft, P Jeurissen, M Faber. Onnodige zorg in de Nederlandse gezondheidszorg, gezien vanuit het perspectief van de huisarts. Celsus academie voor betaalbare zorg. 2013.

K Flobbe, **JJG Wammes**, M Harmsen. Prestatie-indicatoren voor radiologie uit multidisciplinaire richtlijnen. NVVR. 2013.

S Winters, **JJG Wammes**, M Ouwens, M Faber. De toetsing van toetsingsinstrumenten. Evaluatie van toetsingsinstrumenten van patiëntenorganisaties vanuit diverse perspectieven. IQ Healthcare. 2012.

JJG Wammes, M Hilbink-Smolders, T Kool. Versnelling en vernieuwing richtlijnen Jeugdgezondheid. IQ Healthcare. 2012.

English

JJG Wammes, P van der Wees, M Tanke, G Westert, P Jeurissen. A systematic review of high-cost patients' characteristics and healthcare utilization. BMJ Open 2018.

JJG Wammes, M Tanke, W Jonkers, G Westert, P van der Wees, P Jeurissen. Characteristics and healthcare utilization patterns of high-cost beneficiaries in the Netherlands; a cross-sectional claims database study. BMJ Open. 2017.

P van der Wees, **JJG Wammes**, R Akkermans, J Koetsenruijter, G Westert, A van Kampen, G Hannink, M de Waal-Malefijt, W Schreurs. Patient-reported health outcomes after total hip and knee surgery in a Dutch University Hospital Setting: Results of twenty years clinical registry. BMC Musculoskeletal Disorders. 2017.

P van der Wees, **JJG Wammes**, P Jeurissen, G Westert. The role of physicians and their professional bodies in containing healthcare costs. Family practice. 2017.

P van der Wees, **JJG Wammes**, P Jeurissen, G Westert. Stewardship of primary care physicians to contain costs in health care: an international cross-sectional survey. Family practice. 2017.

JJG Wammes, E van den Akker, E Verkerk, S van Dulmen, G Westert, T van Asselt, T Kool. Identifying and prioritizing lower value services from Dutch specialist guidelines and a comparison with the UK do-not-do list. BMC Medicine. 2016.

JJG Wammes, P Jeurissen, G Westert, M Tanke. The Dutch Health Care System 2016. International Profiles of Health Care Systems. The Commonwealth Fund. 2017.

I Mosca, P van der Wees, E Mot, **JJG Wammes**, P Jeurissen. Sustainability of Long-term Care: Puzzling Tasks Ahead for Policy-Makers. International Journal of Health Policy and Management. 2016.

P van der Wees, **JJG Wammes**, G Westert, P Jeurissen. The Relationship Between the Scope of Essential Health Benefits and Statutory Financing: An International Comparison

Across Eight European Countries. International Journal of Health Policy and Management. 2015.

JJG Wammes, P Jeurissen, G Westert. The Dutch Health Care System 2015. International Profiles of Health Care Systems. The Commonwealth Fund. 2015.

JJG Wammes, P Jeurissen, G Westert. The Dutch Health Care System 2014. International Profiles of Health Care Systems. The Commonwealth Fund. 2014.

JJG Wammes, P Jeurissen, G Westert. The Dutch Health Care System 2013. International Profiles of Health Care Systems. The Commonwealth Fund. 2013.

JJG Wammes, W Verstappen, S Gaal, M Wensing, Organisational targets of patient safety improvement programs in primary care; An international web-based survey. BMC Family Practice. 2013.

I Mosca, E Mot, P van der Wees, **JJG Wammes**, Francesca Colombo, P Jeurissen. Universal Challenge: Sustainable Long-Term Care. Celsus academie voor betaalbare zorg. 2013.

C Scholzel-Dorenbos, A Arons, **JJG Wammes**, M Olde-Rikkert, P Krabbe. Validation study of the prototype of disease-specific index measure for health-related quality of life in dementia. Health and Quality of Life Outcomes. 2012.

JJG Wammes, A Siregar, T Hidayat, R Raya, R van Crevel, A van der Ven, R Baltussen. Cost-effectiveness of methadone maintenance therapy as HIV prevention in an Indonesian high-prevalence setting: A mathematical modeling study. The international journal on drug policy. 2012.

A Ganser, G Roth, J van Galen, J Hilderink, **JJG Wammes**, I Muller, F van Leeuwen, K Wiesmuller, R Brock. Diffusion-Driven Device for a High-Resolution Dose-Response Profiling of Combination Chemotherapy. Analytical Chemistry. 2009.