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Systematic review of high-cost patients' characteristics and healthcare utilisation

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ABSTRACT

Objectives To investigate the characteristics and healthcare utilisation 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 30 October 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 utilisation of the top-X% (eg, top-5% and 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 behavioural model was used to categorise 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' utilisation. 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 years. High costs were associated with higher incomes in the USA but with lower incomes elsewhere. Preventable spending was estimated at maximally 10% 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 utilisation 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.

Strengths and limitations of this study

- Based on an extensive literature search, this review included 55 studies of high-cost patients' characteristics and healthcare utilisation.
- Andersen's behavioural model was used to categorise 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 utilisation 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 healthcare 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 USA, many providers or insurance plans have pursued this logic and developed programmes for 'high-need, high-cost patients'. So far, such programmes, 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.[6] Research has shown that the effectiveness and efficiency of the programmes 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-utilisation 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 programme responses.

We conducted this systematic review to synthesise the literature on high-cost patients' characteristics and healthcare utilisation. Andersen's behavioural model (see Methods section) was used to organise 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.

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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 on 30 October 2017. Full details of our search strategy are attached in online supplementary 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 utilisation of the top-X% (eg, top-5% and 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 (eg, hospital costs and 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, inclusion 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 healthcare use and costs (per subpopulation). We also made a narrative summary of the findings per article (provided in online supplementary 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 categorised and presented at the level of International Statistical Classification of Diseases, 10th Revision (ICD-10) chapters.

Data synthesis

Andersen's behavioural model was used to categorise 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 (eg, age, gender, education, ethnicity and beliefs); (2) *enabling* characteristics that facilitate or impede use of services (income/wealth/insurance as ability to pay for services, organisation of service provision and health policy); and (3) *needs* or conditions that laypeople or healthcare providers recognise 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 hypothesised 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]

We presented all data according to five general categories, including study characteristics, predisposing characteristics, enabling characteristics, need characteristics, and expenditure categories and healthcare utilisation. We presented summary tables of results, extracted central themes and topics from the studies and summarised them narratively. All studies were analysed 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.

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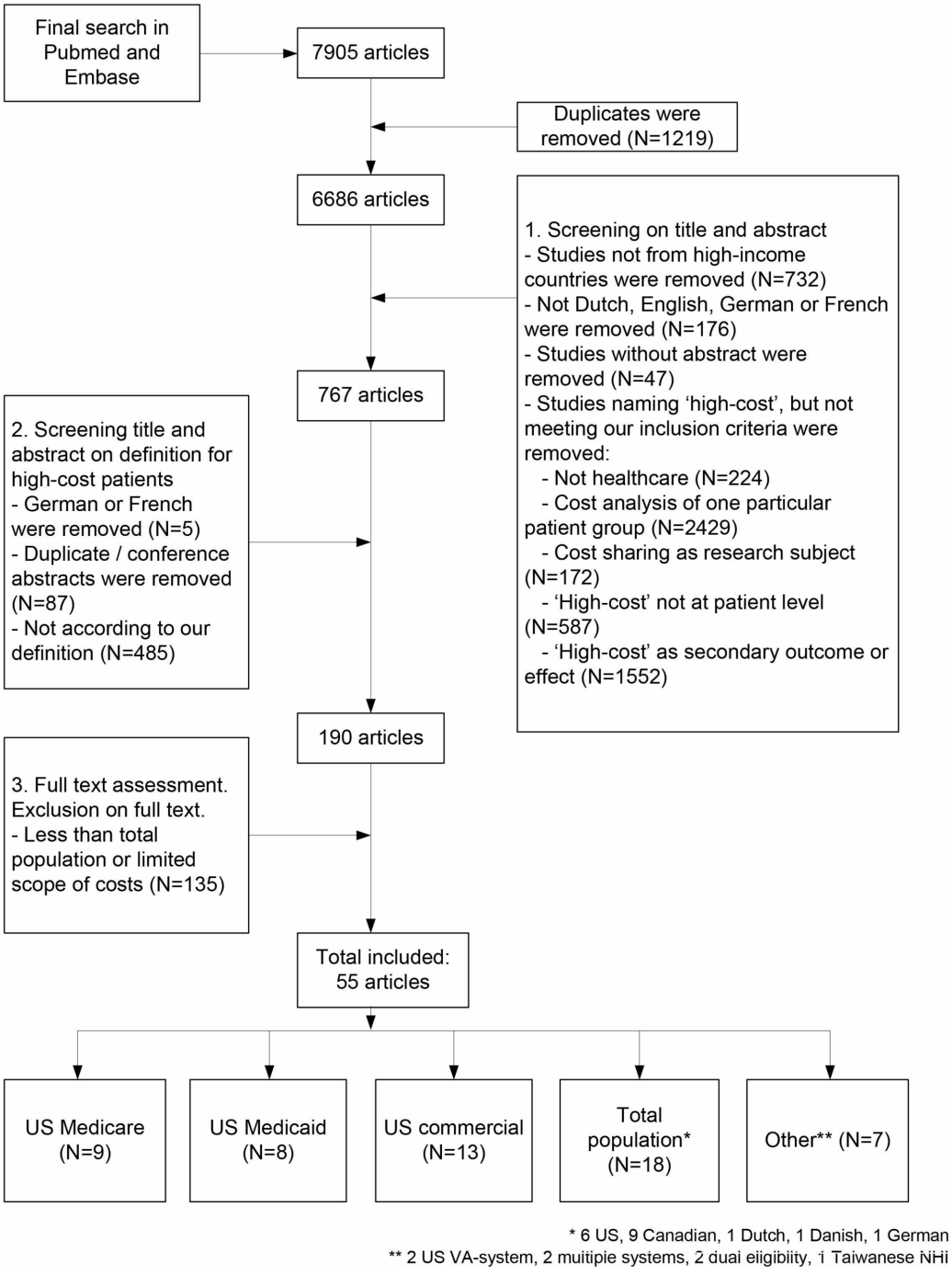


Figure 1 Flow diagram of article selection.

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 USA (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 6 months to 30 years. The most frequent observation period was 1 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-cost 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.

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Table 1 Description of the included studies

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: inclusion and exclusion criteria	Cost data
Aldridge and Kelly,[57] USA	Descriptive	2011	Top-5%	US population	Total spending was identified from a combination of data from Medical Expenditure Panel Survey, the Health and Retirement Study, peer-reviewed literature, published reports, 2011 MEPS and 2011 National Health Expenditure Accounts.
Ash *et al*,[58] USA	Descriptive, logistic regression	1997–1998	Top-0.5% with highest predicted costs, top-0.5% prior cost.	Individuals eligible for at least 1 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*,[59] USA	Predictive modelling, 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*,[28] USA	Descriptive, logistic regression	2011–2012	Top-10%	Fee-For-Service Medicare population. Excluding patients <65 years, enrolled in Medicare advantage and those not continuously enrolled in parts A and B.	Standardised Medicare costs, excluding prescription drug charges.
Boscardin *et al*,[60] USA	Descriptive, logistic regression	2009	Top-10%	Employees enrolled in the Safeway health insurance programme in 2009, with biometric and self-reported health status data (Health Risk Questionnaire). Excluding dependents covered through a family member.	Safeway's health plan.
Buck *et al*,[61] USA	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*,[16] USA	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.
Chang *et al*,[62] USA	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 four health plans who were (1) continuously enrolled, (2) incurred ≥$100 each year, (3) from the 4 largest plans; (4) aged between 18 and 62 years in 2007. Excluding those who died.	Commercial health plans.
Charlson *et al*,[63] USA	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 Medicaid Managed Care costs, including inpatient, outpatient, emergency room, laboratory tests and prescription drugs.
Charlson *et al*,[64] USA	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, behavioural health and prescription drugs.
Chechulin *et al*,[22] Canada	Logistic regression	2007/2008–2010/2011	Top-5%	All Ontario residents serviced by the Ontario healthcare system during the fiscal year 2009/2010. Patients under 5 years or who died during this year were excluded.	Total health system costs (including Long Term Care), excluding outpatient oncology, outpatient dialysis, and outpatient clinic.

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Table 1 Continued

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: inclusion and exclusion criteria	Cost data
Cohen *et al*,[65] USA	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*,[66] USA	Descriptive	2006–2007 (1 year)	Top-10%	Medicare beneficiaries and dual eligibles.	Spending paid for by the public programmes.
Coughlin and Long,[67] USA	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 State Children's Health Insurance Program (SCHIP) coverage or never full benefits. Top-0.1% of spenders.	Medicaid.
Crawford *et al*,[68] USA	Neural network modelling	1999–2001	Top-15%	Members of a health plan, where American Healthways, Inc. provided disease management services. Only members with 24 months continuous enrolment were included.	Health plan costs.
DeLia,[20] USA	Descriptive, multinomial regression	2011–2014	Top-1%, top-2%–10%, Persistently extreme: 4 years top-1% Persistently high: 4 years in top-10%	Medicaid/Children's Health Insurance Program (CHIP) beneficiaries in New Jersey, newly covered individuals under the Affordable Care Act (ACA) (2014) were excluded; Medicaid/Medicare dual eligibles were excluded.	Medicaid FFS claims and managed care encounters and CHIP.
de Oliveira *et al*,[18] Canada	Descriptive	2012	Top-10%, top-5%, top-1%. Mental health HC patients: mental health>50% of total costs.	All adult patients (18 years and older) who had at least one encounter with the Ontario healthcare 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*,[30] USA	Descriptive, χ^2	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 healthcare delivered in the state with the exception of Medicare FFS.
Figueroa *et al*,[39] USA	Descriptive	2012	Top-10%	All Medicare patients, excluding those with Medicare Advantage coverage, who were not continually enrolled in parts A and B.	Standardised Medicare costs.
Fitzpatrick *et al*,[21] Canada	Descriptive, logistic regression	2003/2005 and 5-year follow-up	Top-5%	Participants from two cycles of Canadian Community Health Survey (CCHS) surveys, representative of the population ≥12 years and living in private dwellings. ≥18 years. Excluding baseline high cost.	Ontario health insurance plan.

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Table 1 Continued

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: inclusion and exclusion criteria	Cost data
Fleishmann and Cohen,[69] USA	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*,[23] USA	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.
Graven *et al*,[29] USA	Descriptive	2011–2013	Top-10%, Episodically high-cost, persistently high-cost	Adults ages 19 and over, enrolled in Oregon Medicaid, commercial or Medicare Advantage programmes. Only those with continuous enrolment in 2011 and 2012 were included. Excluding dual eligibles and individuals who had 'coordination of benefit'- laims 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*,[19] 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 5 years.	All publicly funded healthcare in a universal public healthcare system.
Guo *et al*,[36] USA	Descriptive, logistic regression	1999–2000	Top-10% of average monthly expenses	Medicaid, FFS recipients younger than 65 years. Excluding nursing home recipients.	Medicaid costs.
Hartmann *et al*,[70] Germany	Logistic regression	2010–2011	Top-10%	Enrollees 18 years and older of AOK Lower Saxony, Germany's 10th largest statutory health insurer.	Inpatient and outpatient care, sickness benefits, rehabilitation, home nursing, ambulatory drug supply, prescribed therapeutic appliances and remedies.
Hensel *et al*,[71] 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 healthcare, 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*,[72] USA	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 ageing out of dependent status and so on.	Data from all carve-outs (eg, prescription drug and 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 (eg, OTC drugs and travel costs).

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Table 1 Continued

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: inclusion and exclusion criteria	Cost data
Hunter *et al*,[73] USA	Descriptive, linear regression	Fiscal year 2010	Top-5%	Cohort from Veterans Affairs (VA) administrative records, who were eligible for and received care in study period. Excluding individuals with schizophrenia, bipolar depression, other psychosis, alcohol dependence and abuse, drug dependence and abuse, post-traumatic stress disorder and/or depression.	Inpatient, outpatient, pharmacy and non-VA contract care.
Hwang *et al*,[37] USA	Descriptive, logistic regression	2008–2011	Top-10%	Employees from a large employer in Pennsylvania and the employees' dependents. Only those continuously enrolled.	Amount paid by the insurer and the amount of cost sharing paid by individuals.
Izad Shenas *et al*,[74] USA	Data mining techniques/ predictive modelling	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*,[75] USA	Descriptive	2011 and 2012	Top-10%	All Medicare patients, excluding those with Medicare Advantage coverage, who were not continually enrolled in parts A and B, or who died during the study period.	Standardised Medicare costs.
Joynt *et al*,[26] USA	Descriptive, linear regression	2009–2010	Top-10%	Medicare >65 years population. Excluding decedents, any Medicare advantage enrolment, not continuously enrolled.	Inpatient and outpatient services.
Krause *et al*,[76] USA	Logistic regression	2009–2011	Top-5%, top-1%, >$100 000	Enrollees of Blue Cross Blue Shield of Texas, only members 18–63 years, with a zip code in Texas and continuous enrolment in 2009 were included.	Total claims expense, including expenditures for hospital care, outpatient facility services and professional services.
Ku *et al*,[34] Taiwan	Descriptive, generalised estimating equations	2005–2009	Top-10%, top-11%–25%	Survey respondents 65 years of age and older.	National health insurance.
Lauffenburger *et al*,[77] USA	Descriptive, group-based trajectory modelling	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*,[78] USA	Descriptive, cluster analysis	2012	Top-10%	Medicare patients hospitalised exclusively at Cleveland Clinic Health System and received at least 90% of their primary care services at a CCHS facility.	CCHS facility costs, postacute care services were only included for those patients who were admitted to a CCHS postacute care facility.
Leininger *et al*,[79] USA	Descriptive, logistic regression	2009–2010 (1 year)	Top-10%	New enrollees for Medicaid who completed a self-reported health needs assessment.	Medicaid costs.
Lieberman *et al*,[33] USA	Descriptive	1995–1999	Top-5%	Medicare FFS beneficiaries.	Medicare spending.

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Table 1 Continued

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: inclusion and exclusion criteria	Cost data
Meenan *et al*,[80] USA	Risk modelling.	1995–1996	Top-0.5%, top-1%	Enrollees of six Health Maintenance Organizations (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,[31] USA	Descriptive, logistic regression	1996–1997	Various. Top-1%, Top-2%, Top-5%, Top-10%, Top-20%, Top-30%, Top-50%.	Representation of non-institutionalised civilian US population (survey respondents).	Total payments (including Out-Of-Pocket, uncovered services and third-party payments).
Powers and Chaguturu,[9] USA	Descriptive	2014	Top-1%	Patients of Partners HealthCare integrated delivery system.	Medicare, Medicaid, commercial insured populations are compared.
Pritchard *et al*,[35] USA	Descriptive	2011	Top-5%	Managed care population, of all ages, with at least 180 days continuous enrolment prior 1 January 2011, patients with gaps in enrolment greater than 30 days were excluded (so no uninsured or patients enrolled in traditional FFS Medicare or Medicaid programmes).	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*,[38] 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*,[81] Canada	Descriptive	1996–1997 (1 year)	Top-5%	≥18 years and older enrolled in the province's universal healthcare plan.	Medical services costs in a universal healthcare plan (physician and hospital services).
Reschovsky *et al*,[27] USA	Descriptive, logistic regression	2006 or 12 months before death	Top-25%	Medicare FFS beneficiaries, ≥1 Community Tracking Survey survey, with usual source of care physician. Excluding end-stage renal disease beneficiaries.	Standardised total costs of Medicare parts A and B.
Riley,[82] USA	Descriptive	1975–2004	Top-1% Top-5%	Medicare, beneficiaries entitled to parts A and B.	Medicare costs.
Robst,[83] USA	Descriptive, logistic regression	2005–2010	Top-1% in some years, or in 6 years	Medicaid beneficiaries with fee-for-service coverage for at least 6 months in all 6 years.	Medicaid.
Rosella *et al*,[24] 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: institutionalised. Full-time members of the Canadian forces. Persons living in remote areas/aboriginal reserves. Ages 12–18 years.	Those covered by Ontario's Universal Health Insurance Plan. Excluding some prescription drug costs, allied health services, dental care, eye care and assistive devices.
Snider *et al*,[25] USA	Logistic regression	2004–2009	Top-20%	Employees from large US employers, from the Thomson Reuters Marketscan Commercial Claims and Encounters database with both body mass index and claims in any given year. Pregnant women and underweight employees were excluded.	All inpatient, outpatient and prescription claims.
Tamang *et al*,[32] Denmark	Descriptive, prediction modelling	2004–2011	Top-10%	Entire population of Western Denmark, with a full year of active residency in year 1.	Danish National Health Service.

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Table 1 Continued

Author(s), country	Methodological approach	Study period	Definition high-cost	Study population: inclusion and exclusion criteria	Cost data
Wammes *et al*,[17] the 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*,[15] Canada	Descriptive	1 April 2009–31 March 2012	Top-1% Top-5% Top-10% Top-50%	People with a recorded age of less than 105 years who were alive on 1 April in any of the three study years and who had a valid Ontario healthcare at any time between 1 April 2009 and 3 March 2012.	Costs refer to healthcare expenditures that have been allocated to patient encounters for healthcare. All medically necessary care, both acute and long term, as covered by public health insurance. Excluding public health, community service agencies and many other programmes, as well as for administrative (government) staff. Private home care, privately insured medication costs.
Zhao *et al*,[84] USA	Descriptive, linear regression	1997–1999	Top-0.5%	Private insured, whose claims were covered in the Medstat MarketScan Research Database; a multisource private sector healthcare database. All cases with a pharmacy benefit and at least 1 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*,[85] USA	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.

The study population differed between the studies. We categorised 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 9, 7 and 14 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 50 studies, total costs per patient were based on the insurance plan or public programme. In the remaining studies, total costs were based on a survey or identified from a variety of sources.

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.[15] As younger groups are generally healthier, costs are concentrated among fewer individuals. Fourth, clinical diagnoses and utilisation patterns varied across age groups,[15–17] and some subgroups were related to particular ages, including mental health high-cost patients among younger ages.[18] Finally, although age was related to high costs, total population studies showed that approximately half of the high-cost populations were younger than 65 years.[17 19]

Studies showed inconsistent results for gender. Respectively 9 and 16 studies noted males and females were over-represented 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,[17 20] or males in mental health high-cost patients.[18]

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

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

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

Variables	Number of studies
Predisposing factors	
Age	32[17 20–22 24–31 34–36 57 59–62 65 67–72 76 77 79 81 82]
Gender=male	9[17 18 20 22 26 27 36 78 83]
Gender=female	16[17 19 20 24 25 29–31 59–61 65 67 72 75 81]
Ethnicity=black/African–American	4[26–28 82]
Ethnicity=white	5[21 24 61 67 83]
Ethnicity=less likely black or Hispanic	3[31 67 83]
Ethnicity=less likely immigrant	1[21]
Ethnicity=less likely whites	2[75 76]
Region	4[26 67 72 74]
Urban residence	6[19 26 28 34 36 75]
Rural residence	2[22 72]
Living institutionalised	3[20 27 66]
Employment status: early retiree	1[72]
Job satisfaction	1[60]
Marital status: divorced/widow/separated/living alone	2[34 65]
Dependents less likely to incur high costs	1[70]
Receive care in many census divisions	1[27]
Harmful habits	3[24 60 79]
Union membership	1[72]
Education: less than a high-school degree (neighbourhod level)	1[76]
Enabling factors	
Health insurance	
Medicare: more likely dual eligible	6[26–28 39 75 82]
Medicaid: specific eligibility status	4[36 61 67 83]
Commercial: increased insurance	2[59 72]
Total population: insurance status had no effect	1[31]
Type of insurance	1[70]
Income	
Positive relation with high costs	3[31 65 72]
Negative relation	5[18 21 22 71 81]
No relation	3[24 27 34]
Organisational enabling factors	
Primary care physician supply	1[26]
Specialist physician supply	1[26]
Hospital bed supply	1[26]
Medical specialist as usual source of care	1[27]
Proportion of physicians who are medical specialists	2[28 27]
Inadequate time during office visits	1[27]
Proportion of providers operating for profit	2[28 27]
Teaching hospitals	1[28]
Low nurse-to-staffing ratios	1[28]
Low supply of long-term care beds	1[28]

Continued

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Table 2 Continued

Variables	Number of studies
Regular medical doctor or hospital	1[79]
Regular medical doctor (negative relation)	1[24]
Need factors	
A00–B99 Certain infectious and parasitic diseases	9[15 17 20 21 25 62 65 72 83]
C00–D48 Neoplasms	21[15 17 21 22 25 26 28 29 34 35 37 38 72 73 75–78 81 82 85]
D50–D89 Diseases of the blood and blood-forming organs and certain disorders involving the immune mechanism	4[16 20 35 81]
E00–E90 Endocrine, nutritional and metabolic diseases	32[16 17 20–22 25 26 28–30 32–34 36 37 58–60 62 66–68 70 73 75 77–79 81 82 84 85]
F00–F99 Mental and behavioural disorders	34[9 15–18 20–22 24 26 28 29 33 36 38 39 60–63 66 67 70–73 75 77–79 81–83 85]
G00–G99 Diseases of the nervous system	10[17 20 25 32 37 38 62 75 81 83]
H00–H59 Diseases of the eye and adnexa	5[17 21 36 38 81]
I00–I99 Diseases of the circulatory system	36[9 15–18 20–22 26 28 29 32–35 37–39 58–60 62 66 68 70 72 73 75–79 81 82 84 85]
J00–J99 Diseases of the respiratory system	30[9 15–17 20–22 26 28–30 32 34 36–38 58 59 62 65 67 70 73 75 77–79 81 82 84]
K00–K93 Diseases of the digestive system	9[17 18 20 21 38 72 73 81 83]
L00–L99 Diseases of the skin and subcutaneous tissue	5[17 20 21 36 81]
M00–M99 Diseases of the musculoskeletal system and connective tissue	15[9 17 20 21 28 35 60 62 72 73 75 77 78 81 85]
N00–N99 Diseases of the genitourinary system	22[9 16 17 20–22 26 28–30 32 34 35 37 38 70 72 73 75 78 81 82]
O00–O99 Pregnancy, childbirth and the puerperium	5[15 36 39 63 81]
Q00–Q99 Congenital malformations, deformations and chromosomal abnormalities	1[32]
R00–R99 Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	6[17 21 36 60 78 81]
S00–T98 Injury, poisoning and certain other consequences of external causes	9[15 17 21 36 38 72 75 76 78]
Z00–Z99 Factors influencing health status and contact with health services	3[17 21 38]
Chronic illness	22[15 17 20 24 28–30 32–34 36 39 57 59 69 70 73 75 77 81 82 85]
Multimorbidity/burden of comorbid illness	31[9 17 19 20 24–27 29 35 36 39 57 58 60 63 64 68–70 72–75 77 81–85]
Decedents/survival	14[15–17 19 20 27 31–33 36 57 75 81 82]
Activities daily living	7[31 34 59 65 66 69 74]
Health status	9[24 31 33 34 37 59 65 69 74]

in highly deprived or low ethnic concentration neighbourhoods.[21] Other studies found that social deprivation seemed to increase risk for high costs more than material deprivation.[22]

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.[23]

Health behaviours, including underweight, obesity, physical inactivity and former smoking were significantly related to high costs.[24 25]

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

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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 organisation plan, a preferred provider organisation plan or comprehensive insurance compared with high-deductible health plans, and insured status was associated with less consideration of costs in decision making.[23]

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 socioeconomic status (SES) was linked to higher priced care) and also with lower resources use.[23]

Organisational 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.[26] Reschovsky *et al*[27] found several weak or insignificant relationships between organisational 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. 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.[28]

Need characteristics

Medical characteristics of high-cost patients are presented in table 2. We categorised medical characteristics to ICD-10 chapters. Circulatory diseases, mental and behavioural 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*[16] showed that over 26.4% of high-cost US dual eligibles suffered from five or more chronic conditions.

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 29] 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%[30]), 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 with 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 with 11.7% in one US Medicaid study and 5%–13% in total populations. In addition, top-1% patients were more likely to die compared with top-5% patients,[17 31] and persistent high-cost patients were more likely to die than episodic high-cost patients.[32] Finally, among US dual eligibles, mortality varied much across age and residence groups; nearly half of dual eligibles aged 65 years and older died.[16]

Expenditure patterns and healthcare utilisation

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 utilisation (table 3). Inpatient acute hospital care was most often reported as a primary expenditure category for high-cost patients. In line with this, 17 studies reported hospitalisations, admissions or inpatient days as important cost drivers. Lieberman found

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Table 3 Expenditure patterns and utilisation of high-cost patients

Spending category	Number of studies
(Inpatient) hospital care	31[15–19 22–25 27–30 32–39 60 66–68 73 75 78 79 82 85]
Subacute care/postacute care services rehabilitation	11[9 15 22 27 30 35 38 39 66 67 75]
Hospitalisations/admission/patient days/length of stay	17[17–19 23 26 35 36 39 60 68 73 74 77–79 81 85]
Emergency department	12[19 26 29 35–38 60 73 77 78 85]
Outpatient (physician) visits	13[19 27 34–37 39 65 73 77 82 83 85]
Long-term care	11[15 16 22 30 39 66 67 70 73 78 83]
Mental health	10[17 18 22 36 38 61 67 73 83 85]
Physician services	13[15 18 27 35–37 68 73 74 81–83 85]
Intensive care unit	2[78 17]
Prescription drugs	16[17 19 23 30 35–37 62 65 67 68 75 77–79 85]
Persistency	
Subsequent use	13[16 20 21 23 29 31–33 62 67 72 82 83]
Prior use	5[21 32 58 60 65]
Persistent users	21[15 16 20–23 26 29 31–33 37 57 58 60 62 65 67 72 82 83]
Prediction of high-cost patients*	16[22 25 58–60 63–65 68–70 77 79 80 83 84]

*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.

that total spending per beneficiary correlated strongly with the use of inpatient services,[33] likewise several studies found that increasing levels of use (ie, top-1% compared with top-5%) were associated with increasing proportions of spending on (inpatient) hospital care.[15 17 23 24 34 35] Guo *et al*[36] reported that high-cost users consumed more units of each of the service category analysed, with the exception of laboratory tests; these findings were confirmed elsewhere.[35 37] In addition, it was found that 91% of high-cost patients received care in multiple care types.[38] 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 Diagnosis Related Groups >€30 000) contributed to high cost in approximately one-third of top-1% patients and in less than 10% of top-2%–5% patients.[17]

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.[26] 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, chronic obstructive pulmonary disease/asthma and urinary tract infections.[39] Figueroa *et al*[30] found that preventable spending differed by insurance type among US non-elderly: 3.5%, 2.8%, and 1.4% of spending were preventable among US Medicaid, US Medicaid managed care and privately insured high-cost patients, respectively. Similarly, Graven *et al*[29] found that proportions of preventable spending differed between payers and that persistent high-cost patients had higher proportions of preventable spending.

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 with 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.[37]

DISCUSSION

We reviewed 55 studies on high-cost patients' characteristics and healthcare utilisation and made comparisons across payers and countries. The studies consistently point to a high prevalence of multiple (chronic) conditions to explain high-cost patients' utilisation. 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

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contribute to high costs. Preventable spending was estimated at maximally 10% of spending. Furthermore, we found that high costs are associated with increasing age and that clinical diagnoses and utilisation 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 USA, 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 utilisation. 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 USA 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 utilisation of high-cost patients vary across localisations 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 on 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 on 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 on 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 studies 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 40] One contribution of our review is our identification of notable differences in characteristics and utilisation 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 behaviour, which allows for tailored policy.[41] Such segmentation analysis may powerfully inform population health management initiatives. Given the multiple needs and cross-sectoral utilisation of high-cost patients, we suggest such analyses should capture both characteristics and utilisation as broadly as possible, to fully apprehend high-cost patients care needs and utilisation. 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*[42] 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'.

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 organisational factors on spending is in line with Andersen's model predictions, where multimorbidity and health status are prime determinants of healthcare costs.[43] However, such findings are surprising given the abundance of evidence for supplier induced demand and medical practice variation.[44] High-cost populations may be too diverse for studying the impact of organisational 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 10% of spending, which is relatively low compared with 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 programmes, including care coordination and disease management. The algorithms used were said to be relatively narrow and could have included other diagnostic categories.[29] Besides, future studies might consider more broad measures of preventable or wasteful spending and develop algorithms to identify duplicate services, contraindicated care, unnecessary laboratory testing, unnecessary prolonged hospitalisations 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,

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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 characterised 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.[45–47] In addition, health systems might consider strengthening their palliative care systems.[48] 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.[49] In addition, bundled payments for procedures and associated care may improve care coordination and reduce the use of duplicative or unnecessary services.[50] 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.[51 52] 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 53–55] Especially population health management approaches may be beneficial for these populations. Sherry *et al* recently examined five community-oriented programmes that successfully improved care for high-need, high-cost patients. The five programmes shared common attributes, including a 'whole person' orientation, shared leadership, flexible financing and shared cross-system governance structures.[56]

One study addressed health beliefs and patient networks among high-cost patients.[23] More of such research is needed as health beliefs may be more amenable to change than other drivers of high costs. One study analysed the use of expensive treatments by high-cost patients.[17] Better insight in such healthcare utilisation 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 microlevel, mesolevel 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 utilisation 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.

Contributors JJGW drafted the first manuscript and conducted the analyses. JJGW and PJvdW selected eligible studies. JJGW, PJvdW and MACT conceptualised the study and interpreted the data. GPW and PPTJ made a substantial contribution to the development of the research question and interpretation and presentation of the findings. All authors provided feedback to and approved the final manuscript.

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Data sharing statement Detailed forms with extracted data are available from the authors upon request.

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