THE ROLE OF HEALTHCARE DELIVERY, PAYMENT & POLICY INNOVATIONS IN DECREASING THE GLOBAL BURDEN OF CHRONIC DISEASE

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PUBLISHED IN: Frontiers in Public Health

frontiers Research Topics

ISSN 1664-8714
ISBN 978-2-83250-355-3
DOI 10.3389/978-2-83250-355-3

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THE ROLE OF HEALTHCARE DELIVERY, PAYMENT & POLICY INNOVATIONS IN DECREASING THE GLOBAL BURDEN OF CHRONIC DISEASE

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Citation: Howard, S. W., Van De Klundert, J., BeLue, R., eds. (2022). The Role of Healthcare Delivery, Payment & Policy Innovations in Decreasing the Global Burden of Chronic Disease. Lausanne: Frontiers Media SA.
doi: 10.3389/978-2-83250-355-3

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frontiers | Frontiers in Public Health

TYPE Editorial
PUBLISHED 14 September 2022
DOI 10.3389/fpubh.2022.972064

OPEN ACCESS

EDITED AND REVIEWED BY
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Peking University, China

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SPECIALTY SECTION
This article was submitted to Health Economics, a section of the journal Frontiers in Public Health

RECEIVED 17 June 2022
ACCEPTED 22 August 2022
PUBLISHED 14 September 2022

CITATION
Howard SW, BeLue R and Klundert Jvd (2022) Editorial: The role of healthcare delivery, payment and policy innovations in decreasing the global burden of chronic disease. *Front. Public Health* 10:972064. doi: 10.3389/fpubh.2022.972064

Editorial: The role of healthcare delivery, payment and policy innovations in decreasing the global burden of chronic disease

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KEYWORDS

chronic disease, healthcare innovation, health policy, value-base care, alternative payment

Editorial on the Research Topic
The role of healthcare delivery, payment and policy innovations in decreasing the global burden of chronic disease

This Research Topic debuted in 2016 under the title *The Role of Financing, Delivery and Policy Innovations in Decreasing Chronic Disease Burdens*, and has been well received and widely read. In this new Research Topic, we aimed to reprise and refresh the topic of Innovations in Chronic Disease Care, with a renewed focus on value-based and other models of improved care delivery and payment.

The morbidity and mortality caused by chronic diseases increasingly dominates the global burden of disease. Prevention and treatment of these chronic conditions, such as diabetes, hypertension, heart disease, pulmonary conditions, requires the involvement of various formal provider organizations in primary care, specialists and hospitals, as well as of informal care givers. This Research Topic aimed to focus on improvement of health service networks, value-based payment arrangements, and health policies to prevent or more effectively treat chronic diseases. Such improvements often take the form of policy improvements (including financing, regulation, or otherwise), payment and insurance schemes (e.g., insurance, managed care, direct contracting, accountable care, value-based designs), and of interventions in service delivery (care coordination, care processes across organizations, information systems, communication, sharing of resources, etc). Often the most effective improvements involve multi-faceted interventions.

This Research Topic sought to advance the evidence base on effectiveness of interventions to improve the performance of networks servicing patients with chronic conditions, and value-based payment mechanisms aiming to improve outcomes and costs. The medical and health services literature often focuses on interventions addressing single provider organizations, episodes of the full chronic disease patient

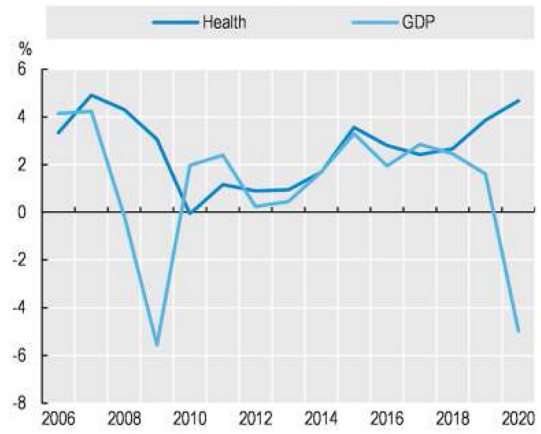

FIGURE 1
Annual real growth in per capita health expenditure and GDP, OECD, 2005–2020. https://www.oecd-ilibrary.org/sites/ae3016b9-en/1/3/7/1/index.html?itemId=/content/publication/ae3016b9-en&_csp_=ca413da5d44587bc56446341952c275e&itemIGO=oecd&itemContentType=book#figure-d1e10418.

journey, and corresponding sub-processes. Less is published on more integrated approaches involving multiple components of the networks, especially research on approaches that include primary and/or secondary prevention. The prevention of onset, early-stage advancement, and subsequent complications and co-morbidities is necessary to manage the societal, organizational, and household-level costs of disease burden.

While addressing chronic disease management and prevention was historically the problem of middle and higher-income countries, improving access to chronic disease-related health care services and reducing the cost of care has become a global imperative. The growing complexity and burden of non-communicable disease, multiple morbidity, and the COVID-19 pandemic have increased demand for care. As a result, costs are increasing due to the rise of the global population, technological advances, and the increasing use of telehealth. When this happens concurrently with plunging GDP in an economic downturn, like in 2008–2009 and 2019–2021, the combined result can be devastating to national economies and public health (Figure 1, OECD, 2022).

In higher-income countries, including the OECD, spending as a percentage of GDP increased from 8.6 to 9.9% from 2010 to 2020 (OECD, 2022). While access in these countries is strong overall, costs require innovative strategies for containment. In many low and middle-income countries, healthcare spending and access lag the needs of the population.

This issue of Frontiers in Public Health features articles on alternative payment strategies and innovations in health care delivery by health services experts from around the world. Several themes converge across the featured articles including the role of reimbursement on health outcomes; the role of consumer knowledge and beliefs and healthcare cost and utilization; utilization patterns in government sponsored insurance programs and novel methods to improve the efficiency and reduce the cost of care for chronic conditions.

The articles selected cover a broad and diverse set of global efforts toward "value improvement" in health care. The original positioning of value-based healthcare in terms of improving outcomes that matter to patients over simply lowering cost is apparent across the studies (1). At the same time, the variety in outcomes and in the financial measures and instruments considered is notable, as is the variety in levels of intervention which range from national policy levels to (sub)organizational contexts. This diversity of policy and organizational context shows how the challenges for value improvement for multiple chronic conditions lie at various levels of the health system and differ across countries and conditions. Each of the studies provides evidence and insights with knowledge and inspiration for value improvement in other contexts. The effectiveness of improved insurance coverage and reimbursement are considered by Xie and Hu and by Ayubcha et al., who report improvements in self-reported health and reduced mortality, respectively. At the same time, Yan et al. provide evidence that such improvements may not be obtained equitably among all targeted patient populations, and more specifically find racial inequities for dementia patients. Reimbursement rates are obviously closely related to out-of-pocket spending, as analyzed in detail for Chronic Disease Syndrome by Close et al. Their analysis reveals the considerable costs involved for individual patients as well as for society at large. Together these financial analyses show the magnitude of the cost of chronic illnesses and how financial policy interventions can improve the outcomes that matter to patients.

Financial incentives might also be targeted to reduce cost and improve outcomes by prevention of illness. This perspective is taken by Salvado et al. who find that the effectiveness of financial incentives for professionals or organizations may vary depending on the type of prevention and incentive scheme. Ranjha et al. and Hosseinejad et al. also focus on efforts to improve the contributions made by professionals. For malaria prevention and elimination, and for community health services, respectively, they identify education and training of the workforce as an important driver of improvement, where Hosseinejad et al. identify a broader bundle of policy interventions needed to strengthen primary care by introducing community health nurses. Interestingly, on the patient side, Ahmad et al. show that education and knowledge may not suffice to produce the desired behaviors and not prevent unnecessary spending.

The studies of Howard et al. and Smeulders et al. focus primarily on outcomes for patients. Smeulders et al. explicitly

distinguish outcomes of general validity and importance from outcomes whose validity is more limited. Such differences in validity of outcomes (and cost) that matter to patients echo recent explicit evidence that invalidate the general applicability of value-based healthcare measures (2). Thus, while the publications in the Research Topics strengthen the knowledge and evidence bases on *The Role of Financing, Delivery and Policy Innovations in Decreasing Chronic Disease Burdens*, they also remind us strongly that value improvement efforts need to be tailored toward the health systems, in particular the financial systems, and need to identify outcomes that matter to the patient in these contexts. The manuscripts published in this Research Topic showcase an inspiring collection of such value improvement research.

Author contributions

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

Acknowledgments

The editors wish to express their appreciation to all authors who submitted manuscripts to this Research Topic, and to Frontiers overall.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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frontiers in Public Health

ORIGINAL RESEARCH
published: 21 August 2020
doi: 10.3389/fpubh.2020.00420

The Economic Impacts of Myalgic Encephalomyelitis/Chronic Fatigue Syndrome in an Australian Cohort

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Objectives: This study aims to estimate direct and indirect health economic costs associated with government and out-of-pocket (OOP) expenditure based on health care service utilization and lost income of participants and carers, as reported by Australian Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS) patient survey participants.

Design: A cost of illness study was conducted to estimate Australian cost data for individuals with a ME/CFS diagnosis as determined by the Canadian Consensus Criteria (CCC), International Consensus Criteria (ICC), and the 1994 CDC Criteria (Fukuda).

Setting and participants: Survey participants identified from a research registry database provided self-report of expenditure associated with ME/CFS related healthcare across a 1-month timeframe between 2017 and 2019.

Main outcome measures: ME/CFS related direct annual government health care costs, OOP health expenditure costs, indirect costs associated with lost income and health care service use patterns.

Results: The mean annual cost of health care related expenditure and associated income loss among survey participants meeting diagnostic criteria for ME/CFS was estimated at $14.5 billion. For direct OOP and Government health care expenditure, high average costs were related to medical practitioner attendance, diagnostics, natural medicines, and device expenditure, with an average attendance of 10.6 referred attendances per annum and 12.1 GP visits per annum related specifically to managing ME/CFS.

Conclusions: The economic impacts of ME/CFS in Australia are significant. Improved understanding of the illness pathology, diagnosis, and management, may reduce costs, improve patient prognosis and decrease the burden of ME/CFS in Australia.

Keywords: myalgic encephalomyelitis, chronic fatigue syndrome, health economics, public health, economic impact, out of pocket cost, health care service utilization, diagnostics

OPEN ACCESS

Edited by:
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Reviewed by:
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Specialty section:
This article was submitted to Health Economics, a section of the journal Frontiers in Public Health

Received: *31 May 2020*
Accepted: *13 July 2020*
Published: *21 August 2020*

Citation:
Close S, Marshall-Gradisnik S, Byrnes J, Smith P, Nghiem S and Staines D (2020) The Economic Impacts of Myalgic Encephalomyelitis/Chronic Fatigue Syndrome in an Australian Cohort. Front. Public Health 8:420. doi: 10.3389/fpubh.2020.00420

INTRODUCTION

Myalgic Encephalomyelitis/Chronic Fatigue Syndrome (ME/CFS), is a debilitating, chronic illness with a high level of social and economic burden due to its disabling, widespread chronic pain, and negative impacts on cognition and multiple body systems (1). The complexity of ME/CFS is compounded by its heterogeneity across onset, symptomatology, relapsing nature, and varying levels of severity ranging from mild impairment to bedridden. ME/CFS often incapacitates individuals over a long period of time, often with a prognosis of increasing severity.

The pathomechanism of ME/CFS is not well-defined. There is no specific laboratory-based diagnostic test and 20 different case definitions have been published (2). Diagnosis relies on assessing patient-reported symptoms to establish whether patients meet a specified case definition, along with extensive testing to exclude other illnesses or causative factors. The use of varying case definition criteria which range from overly broad to highly specified increases the likelihood that persons without ME/CFS are included in clinical trials and interventions distorting clinical trial outcomes and increasing costs for the health care system (1). Understanding the pathology of illness is critical to undertaking robust clinical trials to develop and test diagnostics, treatments, therapeutics and effective clinical management. In the absence of a defined pathology and diagnostic test, it is difficult to establish effective strategies to mitigate the burden and cost of illness.

This study uses three diagnostic criteria; 1994 CDC (Fukuda), Canadian Consensus Criteria (CCC) and International Consensus Criteria (ICC), as outlined in **Table 1**, to ascertain whether participants meet a case definition for ME/CFS. In a recent ME/CFS advisory report, the ICC and the CCC were identified as the most appropriate case definitions and are

TABLE 1 | Case definitions for ME/CFS.

Fukuda (1994 CDC)	2011 International Consensus	Canadian Consensus
REQUIRED PRIMARY SYMPTOM/S		
Chronic debilitating fatigue present for longer than 6 months not relieved by rest, and not due to ongoing exertion	**Post-exertional fatigue** Prolonged, persistent or relapsing, that has been present for longer than 6 months not relieved by rest, and not due to ongoing exertion	**Fatigue, post-exertional malaise and/or fatigue, sleep dysfunction and pain** that persists for at least 6 months
REQUIRED ACCOMPANYING SYMPTOMS		
(4 of the following) Post-exertional malaise Impaired memory or concentration Headaches Muscle pain Joint pain Unrefreshed sleep Sore throat Tender lymph nodes		**Neurological/cognitive** (2 or more of the following) Confusion Impairment of concentration and short-term memory consolidation Disorientation Difficulty with information processing, categorizing and word retrieval -Perceptual and sensory disturbances
	(1 from each of the 4 categories) Neurocognitive Impairment -Difficulty processing information -Short term memory loss Pain/Headaches Sleep disturbance -Disturbed/Unrefreshed sleep Neurosensory, perceptual and motor disturbances: -Neurosensory and perceptual disturbances /Motor disturbances	
	Immune, gastro-intestinal & genitourinary	**Autonomic(a), Neuro endocrine(b) and Immune (c)**
	(1 of the following 5 symptom categories) -Flu-like symptoms -Susceptibility to viral infections with prolonged recovery periods -Gastro-intestinal disturbances -Genitourinary -Sensitivities	(1 of the following from 2 of the 3 categories a, b, c) **(a)** Orthostatic intolerance, neutrally mediated hypotension (NMH), postural orthostatic tachycardia syndrome (POTS), delayed postural hypotension; light-headedness; extreme pallor; nausea and irritable bowel syndrome; urinary frequency/bladder dysfunction; palpitations exertional dyspnea **(b)** Loss of thermostatic stability subnormal body temperature and marked diurnal fluctuation, sweating episodes, recurrent feelings of feverishness and cold extremities; intolerance of extremes of heat and cold; marked weight change anorexia or abnormal appetite; loss of adaptability and worsening of symptoms with stress **(c)** Tender lymph nodes, recurrent sore throat, recurrent flu-like symptoms, general malaise, new sensitivities to food, medications and/or chemicals
	Energy production/transportation	
	(1 of the following 4 symptom categories) -Cardiovascular -Respiratory -Loss of thermostatic stability -Intolerance to extremes of temperature	

recommended for future use. The Fukuda definition, while criticized for being overly broad (3) and potentially resulting in false-positive diagnoses, was included as a diagnostic criterion in this study to allow for comparison with international ME/CFS cost of illness studies (4) and to quantify the costs of participants meeting the Fukuda diagnostic criteria for ME/CFS, as these may be indicative of the costs associated with a misdiagnosis of ME/CFS.

ME/CFS patients are often undiagnosed or experience long delays until diagnosis due to the absence of a lab-based diagnostic test and a lack of General Practitioner (GP) awareness and understanding of ME/CFS (4). In addition to extensive testing, patients will often seek additional and alternative advice and therapeutic options from multiple GPs and a range of health professionals (4). Delays and ambiguity around diagnosis can result in confusion, distress, and poor illness management potentially contributing to patients developing more severe and debilitating forms of ME/CFS (5, 6). Suicide rates are reportedly higher in ME/CFS than comparable conditions (7), likely as a result of the severity and limited options to improve ME/CFS patient quality of life. Improved diagnostic timeframes and clinical management are critical for improving patient outcomes and reducing the burden and economic impact of ME/CFS (4).

No cost-of-illness studies have recently been undertaken in an Australian ME/CFS cohort. Understanding the financial burden associated with ME/CFS and better recognition of the condition will reduce negative impacts on patients and the health care system, progress equitable care for people with ME/CFS, and potentially mitigate high expenditure (1, 8, 9). This study aims to identify epidemiological factors, patient behaviors and expenditure associated with ME/CFS in Australia to improve an understanding of the economic impacts and better inform strategies to mitigate the burden and costs of ME/CFS.

MATERIALS AND METHODS

A cross-sectional economic survey was used to capture indirect and direct costs associated with ME/CFS across 2017–2019. All resource use attributed to ME/CFS was derived using a self-completed online survey. Participants were recruited from the Australian ME/CFS National Center for Neuroimmunology and Emerging Diseases (NCNED) Research Registry Survey database and a research participant network. The NCNED Research Registry Survey database and the research participant network include patients predominantly from across Australia with representation across multiple States and Territories, The registry has been built over the last eight 8 years to capture information about ME/CFS patients and healthy controls, who have participated in NCNED research and trials, have been targeted through ME/CFS advocacy networks or referred by medical professionals post diagnosis of ME/CFS.

Economic survey participant data were matched with participant data from the Research Registry Survey to establish demographic and illness characteristics of patients to enable criteria defined diagnosis of ME/CFS (**Table 2**). The economic survey captured costs directly associated with ME/CFS as identified by participants over a 1-month period immediately prior to completing the economic survey. All data were multiplied by 12 to obtain annual costs (see **Table 3** below).

The study uses a prevalence cost; an aggregate measure of the economic burden of disease reported for a specific time period. It is based on the costs of medical care (direct health system costs), costs associated with accessing care (direct patient costs) and lost income (indirect health care costs). Estimates are based on all individuals diagnosed with or living with the condition (10).

The direct health system costs include hospitalizations, prescription medication, medical devices, diagnostic tests, and attendances with medical and allied health professionals. Direct costs to patients include travel costs, OOP costs for healthcare (i.e., co-payments), non-prescription medicines and formal (i.e., paid) care and support, and insurance premiums. Indirect costs include reduced or lost income patients and carers due to ME/CFS. Cost estimates for prescription, hospitalization and medical services are based on 2019 prices.

Prevalence estimates of ME/CFS in Australia were sourced from a 2013 meta-analysis of prevalence studies (11). ME/CFS diagnostic criteria classification was undertaken using Research Registry Survey responses to ascertain whether participants met the Fukuda, ICC, CCC definitions. Classification analysis was supported by the second author of the paper, who is a specialist in ME/CFS diagnosis and along with methodologies used in other peer-reviewed studies (12). The total annual cost attributed was derived by multiplying the annual cost per person with the prevalence of ME/CFS in Australia. Cost estimates are provided by classification and for the whole ME/CFS population.

For the national cost estimate of the number of people with ME/CFS by specific classification, a hierarchical approach was applied for those who meet multiple classification definitions,

TABLE 2 | Participant characteristics by ME/CFS definition.

Characteristic	Any	FUKUDA	CCC	ICC
n (%)[a]	85 (100%)	18 (21.2%)	23 (27.1%)	44 (51.8%)
Male (%)	24.7%	16.7%	8.7%	36.4%
Indigenous	0.0%	0.0%	0.0%	0.0%
Age (mean)	46.42	51.22	43.22	46.14
Education (%)				
High school	14.1%	16.7%	17.4%	11.4%
Postgrad	32.9%	33.3%	30.4%	34.1%
Professional	20.0%	27.8%	8.7%	22.7%
Undergrad	32.9%	22.2%	43.5%	31.8%
Employment status (%)				
Unemployed	64.7%	72.2%	56.5%	65.9%
Part time	30.6%	27.8%	39.1%	27.3%
Full time	4.7%	0.0%	4.3%	6.8%
Current income (p.a.)	$20,200	$14,281	$17,241	$24,592
Height (cm)	168.50	167.39	165.58	170.48
Weight (Kg)	77.42	84.77	72.36	77.05
BMI	27.35	30.36	26.37	26.62

[a]*Numbers and proportions of participants meeting the Fukuda, CCC and ICC case definitions.*

based on the greater specificity and complexity of ICC and CCC case definitions. Participants meeting these case definitions are more likely to have ME/CFS. Accordingly, all respondents who met the ICC definition (regardless of also meeting other criteria) were considered as ICC and those who met the CCC and Fukuda definition were considered CCC with the remainder just meeting the Fukuda definition being classified as Fukuda. Hence these definitions are not mutually exclusive. Rather, they are overlapping with increasing levels of stringency going from Fukuda to CCC to ICC. However, to aid calculations this approach of simplified proportions was adopted. These simplified proportions of the total ME/CFS population were then applied to the total prevalence estimate of ME/CFS in Australia to estimate the prevalence of each classification.

The cost to the government was estimated based on the price of prescription medications listed on the PBS, less patient co-payment (13). Co-payments applicable to the general population (as opposed to concession card holders) were assumed. The cost of diagnostics and medical attendances was based on the MBS reimbursements (14). OOP costs were reported directly from participants. As patients with ME/CFS are unlikely to meet eligibility requirements for GP chronic disease management plans, attendances with allied health professionals were considered as OOP costs. Productivity costs were derived based on participant self-report of the difference in income pre-onset of illness and income at the time of undertaking the study.

RESULTS

Economic survey respondents were matched with the Research Registry Survey data. Eighty-five of the 163 respondents met one of three definition criteria for ME/CFS. Of the responders meeting a defined case definition, there were more females than males, and the mean age was approximately 46. Majority of responders had a bachelor's degree or postgraduate education (65.8%). 95.3% reported being either unemployed (64.7%) or working part-time (30.6%).

Seventy-eight responders (47.9%) did not meet any of the case definitions. Majority of responders met at least one of the definitions ($n = 85$), with 51.8% of those meeting the most stringent ICC definition ($n = 44$). There was overlap in the classification systems (**Figure 1**) with 36 participants meeting any two definitions, and 15 meeting all three definitions. Using the hierarchical approach to address the overlap between classification systems, 21.2% of participants have been classified under the Fukuda case definition and 27.1 under the CCC case definition.

Previous studies indicated that ICC defined ME/CFS represents a subgroup of ME/CFS patients with decreased physical and social functioning capacity relative to the Fukuda defined groups (6, 15). In this study, all groups reported high levels of unemployment due to illness and substantial indirect costs incurred as a result of lost income. The average annual loss in this cohort was $36,549 for Fukuda, $45,211 for ICC and $55,583 for CCC. There are higher costs and greater losses in income in the ICC and CCC cohorts compared to the Fukuda cohort.

TABLE 3 | Annual service utilization per person by ME/CFS definition.

	Any	FUKUDA	CCC	ICC
Medicines				
Prescription	16.4	16.6	17.2	15.8
Non-prescription and natural medicines	14.8	14.0	14.6	15.3
Total medicines	31.2	30.6	31.8	31.1
Attendances				
Non-referral				
GP	12.1	11.0	12.2	12.5
Nurse	3.4	4.9	3.2	3.0
Total non-referral	*15.6*	*15.8*	*15.4*	*15.5*
Referral				
Neurologist	0.6	0.3	0.6	0.8
Cardiologist	0.7	0.8	0.6	0.8
Gastro-specialist	0.5	0.3	0.6	0.5
Psychologist	4.3	3.3	4.4	4.6
Sleep-specialist	0.8	0.8	0.8	0.8
Pain-specialist	0.4	1.5	0.2	0.0
Radiologist	0.1	0.5	0.0	0.0
Other-specialist	3.1	1.5	3.0	3.8
Total referral	*10.6*	*8.9*	*10.2*	*11.5*
Total attendances	26.1	24.8	25.6	27.0
Devices	7.8	7.7	7.2	8.2
Diagnostics	6.5	4.9	5.0	7.9
Hospitalizations	2.4	2.3	2.2	2.5

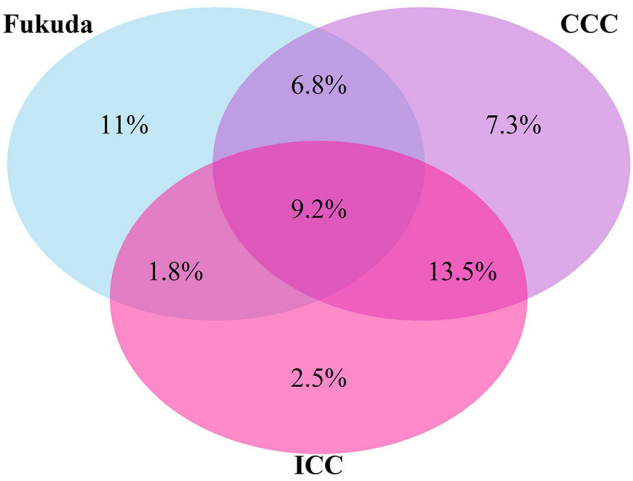

FIGURE 1 | Venn diagram of proportions of participants who met one or more diagnostic Criteria for ME/CFS (%).

TABLE 4 | Annual average per person cost of ME/CFS based on criteria for diagnosis.

Cost	Any	FUKUDA	CCC	ICC
Personal Costs				
Direct health care costs				
Insurance premium	$1,350	$1,280	$1,294	$1,407
Attendances	$1,982	$1,530	$1,858	$2,232
Hospitals	$22	$6	$25	$27
Allied health	$1,115	$1,087	$1,193	$1,085
Diagnostics	$2,343	$1,730	$1,853	$2,848
Prescription medication	$639	$548	$682	$653
Natural Medication	$1,267	$955	$1,217	$1,421
Devices	$8,382	$1,099	$4,148	$13,561
Travel costs	$566	$822	$542	$474
Other costs	$274	$217	$556	$150
Paid support	$600	$752	$598	$540
Total annual average direct out of pocket costs	**$18,540**	**$10,025**	**$13,966**	**$24,398**
Indirect health care costs				
Reduction in Income	$48,757	$36,549	$45,211	$55,583
Reduction in carers income	$3,918	$1,128	$2,825	$5,625
Total annual average indirect out of pocket costs	**$52,675**	**$37,676**	**$48,036**	**$61,208**
Total annual average Personal Cost	$71,215	$47,701	$62,002	$85,606
Government Healthcare Costs				
Community				
Prescription medication	$232	$321	$206	$209
Diagnostics	$683	$488	$639	$785
Attendances	$1,123	$995	$1,110	$1,182
Total community direct healthcare costs	$2,037	$1,803	$1,954	$2,175
Hospital				
Hospitals	$2,445	$2,719	$3,288	$1,893
Total annual average Government Healthcare Costs	**$4,482**	**$4,523**	**$5,242**	**$4,068**
Total Combined Costs				
Total annual average direct health care costs[a]	**$23,022**	**$14,548**	**$19,208**	**$28,466**
Total annual average cost	$75,697	$52,224	$67,244	$89,674

[a] *Total annual average direct health care cost includes government health care costs and direct OOP costs.*

The total average annual cost per person meeting any of the three ME/CFS definitions used in this study is $75,697. Most of the costs were borne by the patient ($71,215), compared to healthcare costs borne by the government ($4,482). Despite infrequent hospitalization (**Table 4**), hospital costs were the largest single cost to governments ($2,445), followed by costs for medical professional attendances ($1,123). Indirect healthcare costs (reduced patients and carers income) was the largest cost ($48,757 and $3,918, respectively) to patients, followed by direct OOP costs associated with devices, diagnostics, and medical professional attendances. The cost to individuals for natural medicines is almost two times that of prescription medication ($1,267 vs. $639).

Using a national prevalence of 0.76% (11), there are an estimated 191,544 Australians living with ME/CFS. The estimated total cost of ME/CFS in Australia was $14,499 million annually (**Table 5**). The estimated cost to the Australian Government was $858 million per annum. Based on 95% confidence intervals of the prevalence estimates, the total cost estimate ranges from $3,335 million to $18,704 million.

TABLE 5 | Estimated total cost of ME/CFS in Australia, 2017–2019.

	Any	FUKUDA	CCC	ICC
Direct OOP costs per person	$18,540	$10,025	$13,966	$24,398
Indirect costs per person	$52,675	$37,676	$48,036	$61,208
Personal cost per person	$71,215	$47,701	$62,002	$85,606
Government cost per person	$4,482	$4,523	$5,242	$4,068
Prevalance[a]	0.76%	21.1%	27.1%	51.8%
Population Estimate (N)	191,544	40,441	51,838	99,265
Direct OOP costs per person	$3,551	$405	$724	$2,422
Indirect costs per person	$10,090	$1,524	$2,490	$6,076
Total personal cost (Mill)	$13,641	$1,929	$3,214	$8,498
Total government cost (Mill)	$858	$183	272	404
Total direct[b] costs (Mill)	**$4,409**	**$588**	**$996**	**$2,826**
Total Cost (Mill)	**$14,499**	**$2,112**	**3,486**	**8,901**
95% LCI	$3,335	$486	802	2,047
95% UCI	$18,704	$2,724	4,497	$11,483

[a]*Fukuda, CCC, ICC and no classification prevalence as a proportion of total prevalence estimate.*
[b]*Total direct cost includes government health care costs and direct OP costs.*

DISCUSSION

This study was undertaken to establish the direct and indirect economic costs associated with ME/CFS in an Australian cohort. It extrapolates those costs to the estimated Australian ME/CFS population to examine the healthcare use profile and types of expenses associated with managing ME/CFS. Of the estimated $14.5 billion annual Australian cost, 70% was due to lost income, 24% due to direct personal OOP costs on health and medical expenditure, and 6% incurred as a cost to government and the health care system.

This cost is significant and is comparable with international studies. A 2011 study in the United States (US), estimated a direct expenditure of USD$14 billion in national healthcare costs and USD$37 billion in lost productivity (16). A 2008 study estimated total direct annual costs in the order of USD$2 billion-9 billion (17) with annual direct costs up to USD$8,854 per ME/CFS patient (17). This study estimates the direct annual health care costs per patient in Australia ($23,022), including direct OOP costs per patient ($18,540) and direct government healthcare costs ($4,482), to be significantly more than the upper US estimates (see **Table 4** above).

The OOP expenditure associated with ME/CFS alone in this study represents a higher proportion of OOP costs spent on health care by participants than by the overall broader Australian population, estimated to be 18% in 2009/10 (18). The average monthly direct personal cost estimated in the study was almost 10 times the estimated direct personal costs incurred (∼$160 per/month) by chronic obstructive pulmonary disease, which represented 3% of the total burden of disease and injury in Australia in 2003 (19). The average direct annual OOP cost estimated in this cohort ($3.5 billion) was estimated to make up 7% of the estimated per capita OOP payments, $24.3 billion, made by Australians between 2011 and 2012 (20).

Understanding cost profiles is important to establish where high expenditure and health service use exists, and how these might influence decision making around support requirements and/or opportunities for cost reductions. This study indicated a high level of expenditure associated with natural medicines, devices and diagnostics, and a broad array of medical and allied health professional attendances. The largest OOP cost relates to devices, and the largest cost to government due to hospitalizations. Compared to the 2018 national average, patients that meet any of the ME/CFS definitions have approximately twice as many visits with a GP (12.1 vs. 6) (21).

High testing costs and medical specialist costs are associated with managing ME/CFS as there are no laboratory-based tests available to diagnose the illness and diagnosis involves testing to exclude other conditions. As the Fukuda definition has been identified as being overly broad, the costs associated with participants in this study meeting the Fukuda definition only, may be indicative of costs associated with misdiagnosis and of not having a lab-based diagnostic test. Based on the estimated prevalence cost of this study, this cost is in the order of two billion dollars.

There is presently insufficient evidence that the use of nutritional supplements and elimination or modified diets relieves ME/CFS symptoms (22). Despite this expenditure on supplements was high. In the absence of clinical evidence of effective treatments for ME/CFS (19), medications and supplements that do not alleviate symptoms of individual patients may be an unnecessary expense. Training and educating health professionals to diagnose and provide appropriate treatment may improve patient prognosis and reduce higher costs associated with more severe ME/CFS (5).

High OOP costs for long term chronic illness patients are of increasing concern to the Australian health system and society as the financial strain on individuals can result in individuals not seeking adequate health care and their condition worsening (23). This is particularly concerning for ME/CFS patients who may further avoid seeking out health care due to low expectations around receiving adequate care and support (24).

Estimated extrapolated costs in the study population are exceedingly high for a relatively small percentage of the population. Considering the low federally funded expenditure on ME/CFS research in Australia to date, there is potential to significantly reduce the public health burden of ME/CFS in Australia. Better understanding, diagnosis, treatment and management of the illness, and better support for patients and carers will be critical to reducing such costs (1, 8). A laboratory-based diagnostic test has further potential to significantly decrease costs by providing greater certainty in identifying ME/CFS patients, undertaking clinical trials, and developing appropriate treatments with proven effectiveness.

The economic impacts of ME/CFS in Australia are substantial for patients and the government. Federal ME/CFS research funding expenditure in Australia is not reflective of the significant economic impacts of the illness. The ongoing and potentially increasing financial burden will be difficult to alleviate without targeted research into the pathomechanism of ME/CFS, development of effective treatment options and improved

diagnosis and management of ME/CFS patients (3). This study provides an indication that the high costs associated with ME/CFS could be significantly reduced through the development of a lab-based diagnostic, more effective treatment options and better management strategies through improved awareness and training for General and Specialist Practitioners.

LIMITATIONS

In this study diagnosis, ME/CFS classification, attribution, and estimates of costs associated with ME/CFS were based on self-reported responses to an online survey. This method is common, despite associated known weaknesses such as recall bias. This may result in classification error, underestimate total resource consumption, and over represent significant events (such as hospitalizations) relative to minor occurrences such as medical attendances. It is unknown if this would over or underestimate the extent to which the reported healthcare use is due to ME/CFS nor what the extent of misdiagnosis might be. The sample was, however, based on participants identified through the ME/CFS research registry survey database, many of whom had received a diagnosis from a health care clinician, with validated diagnostic questions that provide a robust means for classification.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Griffith University Human Research Ethics Committee (GUHREC reference number 2016/502). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

SC, SM-G, JB, and DS contributed to conception and design of the study. SM-G and DS set up and organized the database. SC, JB, and SN performed the data and statistical analysis. SC, JB, and SN wrote the first draft of the manuscript. SC, SM-G, JB, PS, SN, and DS wrote and significantly revised sections of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

FUNDING

This study was supported by the Mason Foundation, McCusker Charitable Foundation, Stafford Fox Medical Research Foundation, Mr. Douglas Stutt, Alison Hunter Memorial Foundation, Buxton Foundation, Blake Beckett Trust, Henty Donation, and the Change for ME Charity.

ACKNOWLEDGMENTS

This study survey was developed by the corresponding author based on the TORPEDO-CF Health Service Diary listed on the Database of Instruments for Resource Use Measurement website (25). The authors would like to acknowledge the contributions of Prof. Elizabeth Unger, Dr. Majella Murphy, Ms. Karen Close, and members of the NCNED team who provided support, guidance, and advice in regard to the paper and survey development, their support is greatly appreciated.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Public Health

ORIGINAL RESEARCH
published: 11 February 2021
doi: 10.3389/fpubh.2021.623966

Kidney Exchange Program Reporting Standards: Evidence-Based Consensus From Europe

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Background: Kidney Exchange Programs can play an important role to increase access to the life saving and most cost-effective treatment for End Stage Renal Disease. The rise of national KEPs in Europe brings a need for standardized performance reporting to facilitate the development of an international evidence base on program practices.

Methods: We systematically searched and reviewed the literature to extract kidney exchange program performance measures. Reported measures were initially categorized as structure, process, and outcome measures. Expert feedback was used to redefine categories and extend the set of measures to be considered. Using the Delphi method and a panel of 10 experts, the resulting measures were subsequently classified as mandatory (Base set), optional (Extended set), or deleted.

Results: Out of the initial 1,668 articles identified by systematic literature search, 21 European publications on kidney exchange programs were included to collect performance measures, accompanied by three national program reports. The final measurement categories were Context, Population, Enrollment, Matching, Transplantation, and Outcomes. The set of performance measures resulting from the literature review was modified and classified as mandatory or optional. The resulting Base set and Extended set form the kidney exchange program reporting standard.

Conclusions: The evidence-based and consensus-based kidney exchange program reporting standard can harmonize practical and scientific reporting on kidney exchange programs, thus facilitating the advancement of national programs. In addition, the kidney exchange program reporting standard can promote and align cross-national programs.

Keywords: kidney exchange, Living donation, reporting standard, end stage renal disease (ESRD), kidney exchange program

OPEN ACCESS

Edited by:
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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *30 October 2020*
Accepted: *15 January 2021*
Published: *11 February 2021*

Citation:
Smeulders B, Mankowski MA and van de Klundert J (2021) Kidney Exchange Program Reporting Standards: Evidence-Based Consensus From Europe. Front. Public Health 9:623966. doi: 10.3389/fpubh.2021.623966

INTRODUCTION

With a mortality number of 1.2 million, Chronic Kidney Disease (CKD) is the 11th most common cause of death globally, and ranks 13th in Europe (1). It is a progressive disease of which End Stage Renal Disease (ESRD) is the last stage. The relative contribution of ESRD to European mortality is increasing, and currently stands at 1.58% in Europe, while CKD accounts for 1.06% of the total burden of disease in Europe (1). The default treatment for ESRD is dialysis (2).

Dialysis incurs higher costs and lower quality of life than transplantation (2). In many European countries, transplantation programs have emanated from deceased donor programs. They have been complemented by living donor programs to promote access and quality of transplantation (3, 4). Live donation traditionally has been restricted to family members or close friends of a patient donating one of their two kidneys. Unfortunately, even when a living donor is available, transplantation may not be feasible because of incompatibility between the patient and the donor.

Over the past two decades, Kidney Exchange Programs (KEPs) have emerged in many countries to promote the benefits of living donor kidney donation. They particularly service pairs consisting of a patient and a living donor willing to donate to the patient for whom transplant is not feasible because the donor is not compatible with the patient. The KEPs "exchange" donors among patients so that patients are matched with compatible donors after which corresponding transplants take place.

Across the world, the design and developments of KEPs have varied considerably. The variations often are solutions to resolve country specific challenges such as small population size (Iceland) geography (Australia) and pre-existing deceased donor programs (Spain) (3, 4). In addition, the variations have arisen from challenges posed by differences in legislation. For example, countries such as Finland and Germany legally forbid living donation to recipients with whom the donor doesn't have a close relationship, whereas in other countries, such as France and Switzerland, altruistic donation is not legally allowed (3, 4).

Another main difference among KEPs arises from differences in national governance models. In many countries, national governance is limited to providing regulation for KEPs. The regulatory frameworks then may govern single center KEPS, such as in the Czech Republic and Slovakia, or KEPs between a small number of centers, such as in Poland. In the USA, less than half of the 250 living donor transplant centers participate in the nationwide KEP administered by UNOS, the organization which manages the nation's organ transplant system under contract with the federal government. In addition, the National Kidney Registry and the Alliance for Paired Donation operate nationwide and regional and single-center KEPs exist. At the other end of the governance spectrum, a national KEP has naturally emerged in the UK with its nationwide public health system. We refer to (3, 5) for a more detailed discussion of KEPS in Europe and across the globe, including in Australia, Canada and South Korea.

The dynamics of the emergent KEPs and their contextual differences have not only resulted in a variety of KEPs but have also brought along a variety of KEP performance measures reported. While this variety of reporting practices promotes novel approaches and viewpoints, it hampers comparability of practices and performance, and ultimately the development of an evidence base on KEP performance, as is beneficial for existing and newly emerging KEPs and most importantly for patients suffering from ESRD. Our research aims to synthesize reported measures and develop a consensus-based set of reporting standards.

Our research focusses on reporting for national KEPs. Moreover, we limit the scope to European KEPs. The reasons for limiting the scope to nationally coordinated KEPs in Europe are 2-fold. First, it serves to limit contextual differences, which complicate consensus and harmonization of standards. European countries and health systems differ essentially from other countries reporting on KEPS, such as Australia, China, India, Iran, Japan, Korea, and the USA, which translate to differences in KEPS and in KEP reporting priorities and practices. Second, Europe is presently witnessing various initiatives for cross-national KEPs (6), which especially call for harmonization of performance measures and reporting among European KEPs. These initiatives include bilateral KEPs between two countries, such as the Czech-Austrian kidney exchange (6) or KEPs involving multiple countries such as the Scandia Transplant Kidney Exchange Program (3).

To accomplish our research aim, we address two research questions. First, we seek to comprehensively map present scientific and practical reporting by European national KEPs. On this basis, we then proceed to develop unified reporting standards for European KEPs. The resulting Kidney Exchange Program Reporting Standards (KEPREPS) consist of a Base Set of essential, mandatory measures and an Extended set of optional measures, valuable to report for some programs. The presented work is developed within the EU COST Action 15210 entitled European Network for Collaboration on Kidney Exchange Programs (ENCKEP).

METHODS

The method section contains two main parts. First is a systematic review and synthesis of European literature on Kidney Exchange Programs. The results of this literature review served as the starting point for the second stage of developing a reporting standard. In this stage, we solicited and processed several rounds of expert feedback following the Delphi method to derive consensus on the Kidney Exchange Program Reporting Standards (KEPREPS) reporting standard. The details of both methods are specified below.

Systematic Literature Review

Data on KEP performance measures were extracted from peer-reviewed English language scientific journals and conference proceedings and (annual) reports from Europe's three largest kidney exchange programs: The Netherlands, Spain, and The United Kingdom. A publication was considered 'European' if it reported data from a European KEP or if the first author was affiliated with a European institution.

After extensive consultation with an expert librarian, we included articles from Embase, Medline, Web of Science, and Cochrane matching the following query:

[(kidney OR donor OR transplant* OR graft*) AND (exchange* OR pair*-exchange* OR pair*-donation* OR sharing)]*
OR
[(pair-exchange* OR pair*-donation* OR ((exchange* OR sharing OR chain) AND (donor* OR donation* OR kidney*))] AND (renal* OR kidney*)]*

In the first round, the first and third author screened the title and abstract of all articles and excluded the articles for which both agreed on exclusion. In a second round, the same two authors considered the full texts of the non-excluded articles, again deciding to exclude only if both authors agreed on exclusion. Differences in assessment on exclusion were resolved by consensus.

Next, all included articles were screened by the same two authors to collect all performance measures reported. Again, the two authors ensured full consensus on each of the reported measures for each of the included articles. Lastly, the first author included all performance measures explicitly included in the annual reports of the British, Dutch, and Spanish KEPs.

Measure Categories

The measures extracted from the literature were subsequently categorized from two perspectives. Firstly, we distinguished different types of measures, using Donabedian's seminal Structure-Process-Outcome framework (7). This resulted in three initial categories of KEP measures-structure measures, process measures, and outcome measures. A fourth set of measures on the population participating in the KEP was added to ensure all measures were categorized. The second perspective regarded the frequency of reporting, which we interpreted to indicate the relevance of the measures. Hence, based on the reporting frequency, we provided an initial classification of measures. Commonly reported measures were initially classified as mandatory, less commonly, but still regularly reported measures were initially classified as optional, and the remainder as exceptional.

Base Set and Extended Set

The systematic review results were presented to the large group of ENCKEP participants, representing 28 predominantly European countries. This large audience of representatives provided feedback and proposed additional measures.

Based on this initial expert feedback, the categories were redefined and formed the input for a procedure following the Delphi method with a set of 10 volunteering expert participants of ENCKEP (8). The experts in the Delphi panel responded to two questionnaire rounds. During the first questionnaire round, the experts categorized measures into three groups: Base set, Extended set, and Other. The Base Set consisted of measures that should be reported by every KEP. The role of the Extended set was to incorporate important but non-essential measures, while the category "Other" accommodated non-essential measures to be excluded from the standard.

The experts were also given the opportunity to provide motivation for their answers and make additional comments. Based on the results of the first round, we calculated the average score for each measure granting 3/2/1 points for Base Set/Extended Set/Other and rounded the score for each measure to the nearest integer. This rounded score was then converted into an initial classification of each measure into Base set (score $\geq$ 2.5), Extended set (2.5 $\geq$ score $\geq$ 1.5), or Other (score <1.5).

In the second round, we presented the resulting Base set and Extended set and asked each expert to agree or disagree. The experts were requested to motivate any disagreement to facilitate further improvement or finalize decision making on the classification. A third round would have been conducted in case of a lack of consensus but was not necessary. The resulting classification formed the targeted reporting standard KEPREPS.

TABLE 1 | List of included European publications and their categorization.

	Publication type	References
Observational	KEP reports	(10–12),
	Journal articles	(13–25)
Model	Real data	(9, 26–29)
	Simulated data	(30–32)

RESULTS: SYSTEMATIC LITERATURE REVIEW

The systematic literature search yielded 1,668 articles. After scanning of titles and abstracts, 346 were included. After reading of the full text (thus eliminating conference papers and posters), 116 articles remained, 21 of which were regarded as European. To this set, we added three publicly available annual reports of major European KEP programs.

The review distinguishes observational empirical studies and (simulation) model studies. No controlled experimental studies have been reported. There are 16 observational studies, including three recent annual reports from the aforementioned KEPs, and 8 model studies. With one exception (9), the observational studies are quantitative studies. The (simulation) model studies used real-life data, generated data, or a combination of both. **Table 1** categorizes the included publications.

The majority of the included studies originate in the Netherlands (13/24) and consider the long-running Dutch KEP. By contrast, only 2 UK documents are included, despite the large size of the UK program. Seven publications are from 2005 to 2009, 9 from 2010 to 2014, the remaining eight from 2015 onwards. Detailed information per included article, including the reported measures, is available as **Supplementary Material**.

Population Measures

Population measures are the measures describing the donors and patients involved in the kidney exchange program. This data can be further divided into three main categories, information regarding program size, demographic data, and medical data.

Program Size

Program size measures are commonly reported. For instance, 12 out of 16 observational studies report the number of patients participating in the KEP, as do seven out of eight model studies. Of the model studies, five publications report on computational challenges in relation to program size. Papers not reporting on program size typically have a specific, different focus, such as validation of virtual crossmatch procedures (9) or KEP transplant outcomes compared to living-related transplant outcomes (13).

Demographics

Demographic data is commonly reported in the observational literature. Age (10/16), the relationship between donor and patient (8/16) and gender (5/16) information is provided often, mainly in papers describing functioning KEP programs. Ethnicity is only mentioned once (14).

In the simulation literature, demographic data is nearly absent. Within this literature, population data is focused on characteristics with a direct impact on the kidney exchange graph.

Clinical Population Measures

The composition of the KEP pool with regards to ABO and immunological characteristics can have a large impact on the overall and individual outcomes. These measures also shed light on patient enrolment causes.

In the observational literature, the type of incompatibility (ABOi, positive crossmatch) within a pair is reported in 11 out of 16 papers. ABO information for donors or candidates (7/16) and patient PRA (8/16) are also commonly reported. In some cases, this information is given by the type of incompatibility. ABO information for patients is often limited to the number of blood type O patients. Reporting on incompatibility types is often in combination with subsequent reporting on outcomes, e.g., transplant probability per incompatibility type (see below).

Simulation papers have less commonly reported on the ABO and PRA typing (3/8) and type of incompatibility (2/8) explicitly. However, an additional two papers refer to the data simulator they employ, which addressed these measures too. **Table 2** summarizes the reporting of population measures.

Structure Measures

There is little reporting on structural characteristics. Five (observational) studies report on the number of transplant centers involved. Four studies report on the spread of transplants over the transplant centers, three of which are model studies.

Process Measures

As depicted in **Table 2**, five studies report on enrolment rates, only one of which is a model study. For the matching process, ten studies report on cycle lengths and numbers before HLA crossmatching, six of which are observational. This topic may have received more interest from model studies because cycle length received considerable attention in the scientific literature in relation to computational complexity. Likewise, three of the five studies which report the length of the longest cycle and/or chain are model studies.

The process (outcome) measure receiving the most attention is the number of transplants. As much as 13 of the 18 observational studies and five out of eight model studies report the total number of transplants proceeding. Seven of these studies distinguished ABO incompatible and crossmatch incompatible pairs, one of which was a model study. Six studies reported transplants per blood type. Two studies reported on the number of blood type identical transplants, and one study reported on ABO incompatible transplants.

TABLE 2 | Reporting of, population, structural, process, and reporting measures by type of study.

		Observational	Simulation
Population measures			
	Number of documents	16	8
Program size	# Patients or pairs	12	8
	# Non-directed donors	2	0
Demographics	Gender	6	1
	Age	11	2
	Ethnicity	1	0
	Pair relationship	9	0
Medical	PRA level	9	4
	ABO	8	4
	Pair incompatibility	11	3
Structural and process measures			
	Number of documents	16	8
KEP structure	Number of centers	5	0
	Enrolment rate	4	1
	Pairs per run	6	2
Match before crossmatch	Identified cycles (by length)	6	4
	Length of longest cycle/chain	2	3
	Patients matched	5	7
Process outcome	Transplants canceled due to crossmatch	4	2
	Transplants proceeding	13	5
	Transplants proceeding by incompatibility	6	1
	Transplants proceeding by ABO	6	2
	Average time to match	3	3
	Patients remaining in pool	6	1
	Abandonment rate	6	1
	Transplants outside KEP	7	0
Others	Cold ischemia time	2	0
	Computation time	0	3
	Transplant increase over other programs	2	0
Reporting measures			
	Number of documents	16	8
Transplant outcomes	Graft survival	5	0
	Patient survival	4	0
	Acute rejection rate	3	0
Descriptive statistics matched patients	PRA	5	0
	Age and gender	3	0
	Patient ABO	3	0
	Relationship to donor	2	0
	Incompatibility to donor	1	0
	Ethnicity	1	0
Descriptive statistics unmatched patients	PRA	3	0
	Age and gender	2	0
	Patient ABO	2	0
	Relationship to donor	2	0
Descriptive statistics matched donor	ABO	2	0

A closely related process measure is the number of matched patients before crossmatch. This measure is reported by 12 studies and by all but one of the model studies. A next closely related measure is the number of transplants canceled because of negative crossmatch. This measure is reported by six studies, only two of which are model studies.

Eight studies (of which six observational) report the average number of patients included per match run, and six studies (of which three are observational) report the average time until being matched. This average can be the overall average but may also distinguish blood types and highly sensitized patients. Seven studies report on the number of pairs remaining in the pool, of which six are observational. The same set of seven studies also reported on the abandonment rate. Seven observational studies reported on the number of patients who received a transplant outside of the exchange program. Several other less frequently reported measures are provided in **Table 2**.

Outcome Measures

In comparison to process or population measures, outcome measures have received less attention. The most frequently reported outcome measures are graft survival rate (five observational studies), patient survival rate (four observational studies), and acute rejection rates (three observational studies). The only qualitative study included reported on psychological outcomes, such as psychological distress and complaints, and the need for support.

Five observational studies-four of which are Dutch-report on descriptive statistics for matched and unmatched patients. All these studies report on PRA levels of matched patients and four for unmatched patients. The age and gender of donors and recipients (matched patients), as well as recipient ABO type, are reported by three studies. Less frequently reported outcome measures can be found in **Table 2**.

RESULTS: TOWARD A REPORTING STANDARD

The above results were discussed with a broad expert panel of ENCKEP participants who proposed additional measures. The discussion led to a revised categorization. The three resulting main categories are: Context Information, Process Measures, and Outcome Measures. The Context Information is subdivided into measures on the program, on participating individuals (recipients and donors) and on pairs. The process measures are partitioned into three sequential subcategories: Enrolment, Matching, and Transplantation.

For each of the categories, measures were classified as essential (Base Set), important but non-essential (Extended Set), or not important (Other) by a Delphi panel of 10 experts from France, Hungary, Italy, The Netherlands, Portugal, Spain, Switzerland, and United Kingdom. In a second Delphi round with the same panel (except one expert), the averaged classifications (see methods section) were proposed to the panel for approval.

The resulting classification, as presented in **Tables 3**, **4**, was approved nearly unanimously. We refer to the **Appendix** for a total of 16 exceptions and expert reservations. Sometimes, disagreement or reservation was because of legal considerations (collecting data on ethnicity is not allowed) or regulatory conventions (reporting measures for the living donor program as a whole). Five of the measures involved were from the Base Set. In each of these cases, one expert disagreed. For varying reasons, the corresponding items were kept in the Base Set. For the items in the Extended Set, we judged minor disagreement was not problematic as reporting of items in the Extended Set is optional anyway.

Table 3 shows the resulting Base Set. Among the added context measures is the definition of incompatibility, considered as necessary context information to interpret reporting on other measures. From the European perspective, participant participation in the Eurotransplant Acceptable Mismatch Program (33) (or similar program for highly sensitized candidates) was also considered essential. For recipient and donor attributes, the Base Set contained blood type, gender, cPRA, and age. Experts also judged reporting on relationship and type of incompatibility for enrolled pairs to be mandatory.

The Base Set process and outcome measures are presented in **Table 3** Experts classified all proposed matching measures as essential (Base Set) except for computation time. In addition to the measures from the review, measures on KEP transplants as a percentage of the total living donor program, and the total increase in donation caused by the KEP are included in the Base Set. While not frequently reported in the systematic review, outcome performance measures on patient, donor, and graft survival were also selected as Base Set measures.

Toward a Guideline on Reporting: Extended Set

Table 4 shows all context information, process, and outcome measures considered to be important but not essential and, therefore, to be included in the Extended Set. Most of the context information, including MFI thresholds, matching algorithm, organ/donor travel, and cPRA definitions, ended up in the Extended Set. The same goes for nationality, ethnicity, social demographics, donor's LKDPI (34), and recipient's match probability. Many of these measures were not in the systematic literature review results. The Extended Set also contained additional outcome measures relating to quality-adjusted life years (QALYs) for donors/recipients, number of rejections, and cost measures.

DISCUSSION

The results presented above provide a Reporting Standard for European KEPs based on systematic literature review and expert opinion collected from a panel of practitioners and scientists from a variety of European countries and KEPs. The literature review made clear that not all existing European KEPs have reported on performance in the scientific literature. Moreover, it is remarkable that more than half of the publications are from The Netherlands, as the UK and Spain have larger KEPs but only

TABLE 3 | Base set of context information, donor/recipient/pair attributes, and performance measures suggested to report by every KEP.

Measure name	Definition	Proposed format
Context information		
Definition incompatibility	Complete (in)compatibility definition used in the KEP	Narrative
Acceptable mismatch possible in KEP	Program for patients qualifying for a 'mismatched donor (cd. eurotransplant acceptable mismatch program)	Narrative
Recipient and donor attributes		
Blood Type	Recipient and donor blood type	Distribution
cPRA	Recipient calculated panel reactive antibodies	Average value/distribution
Prior Transplants	Recipient prior transplants	Number/percentage with prior transplant
Gender	Recipient and donor age	Distribution/average
Age	Recipient and donor gender	Total/percentage per gender
Type of incompatibility	Pair's type of incompatibility (ABO, positive crossmatch)	Percentage among categories/narrative
Relationship	Relationship between donor and incompatible recipient	Percentage among categories/narrative
Enrolment performance measures		
Number of pairs	Number of pairs registered in the KEP	(Total/average) over the reporting period
Number of non-directed donors	Number of non-directed donors registered in the KEP	(Total/average) over the reporting period
Matching performance measures		
Pairs matched	Total number of matched pairs	(Total/average) over the reporting period
Pairs left unmatched	Total number of unmatched pairs	Total/average over the reporting period (e.g., remaining at the end of the reporting period)
Identified cycles/chains (by length)	Cycles and chains found in the matching, by number of patients in the cycle/chain	Total/average over the reporting period, by cycle/chain length.
Number of match failures	Total number of matches found that have failed	Total/average number across the reporting period
Reasons for match failures	Synthesis on match failures reasons	Narrative including common causes
HLA matching	Degree of mismatch in donor and candidate HLA profile	Average value (0–6)/minimium, average, maximum/distribution
Age matching	Age gap between donor and recipient	Average value/minimium, average, maximum/distribution
Transplant performance measures		
Pairs transplanted	Number of pairs transplanted	Total average number across the reporting period
Pairs not transplanted	Number of pairs not transplanted since enrolled	Total/average number across the reporting period (e.g., remaining at the end of period)
Pairs transplanted outside of KEP	Number of pairs enrolled in the KEP but transplanted outside the program	Total number across the reporting period
Average time to transplant	Time from the enrolment to transplant	Average value over the period
Non-directed donor utilization	Number of donations from non-directed donors	Total/average number across the reporting period, ratio
Increase in the total number of transplants due to KEP	Increase in the total number of kidney transplants due to KEP (all living and deceased donation)	Total/average number across the reporting period/rate
Percentage of KEP transplants as part of the total living donor program	Percentage of KEP transplants as part of the total living donor program	(Average) Percentage across the reporting period
Outcome measures		
Patient Survival	Living donor transplant recipients survivors as a percentage of KEP transplant recipients	(Average) Percentage
Donor Survival	Living donors surviving as a percentage of KEP transplant donors	(Average) Percentage
Graft Survival	Living donor transplant grafts surviving as a percentage of KEP transplanted grafts	(Average) Percentage

modestly contributed to the European evidence base in the form of peer-reviewed scientific publications.

Living donor transplantation, as promoted by KEPs, is evidenced to be the most cost-effective treatment for End Stage Renal Disease (35–37). Given the importance of cost-effectiveness in current health policy and decision making, the lack of KEP reporting on cost measures is remarkable, as is the fact that outcome measures (effects) are among the least reported. However, measures on cost and effects (outcomes) have been included in the Base Set of KEPREPS upon expert

TABLE 4 | Extended Set of context information, donor/recipient/pair attributes, and performance measures to report by KEPs.

Measure name	Definition	Proposed format
Context information		
MFI–threshold for (in)compatibility	Mean fluorescence intensity (MFI) level thresholds defining (in)compatibility criteria	Narrative
Matching algorithm	Matching algorithms used in the KEP	Description of algorithmic principles
Prevalence of need for a kidney transplant	National/regional population incidence and prevalence of ESRD	Incidence and prevalence numbers
Donor or organs travel	Distance that donor or organ traveled for transplant	Narrative
Definition cPRA	Exact calculation definition of cPRA	Descriptive
Other programs–where is the KEP in the system of other transplant programs.	Environment of other KEPs, living and deceased donation programs	Narrative
Outcomes of alternate transplant programs	Outcome measures of competing KEPs, living and deceased donation programs	Descriptive
Recipient and donor attributes		
Nationality	Recipient and donor nationalities	Number/percentage per country
Ethnicity	Recipient and donor ethnicity	Number/percentage ethnicity
Social demographics	Recipient and donor socio-economic information expressed statistically, also including employment, education, income	Distribution/rate
Recipient attributes		
Match probability	Recipient match probability within a KEP	Average value/distribution
Donor attributes		
LKDPI	Donor living donor kidney transplantation index	Average value distribution
Matching performance measures		
Computation time	Run-time of algorithms required to output matching pairs	Average value
Transplant performance measures		
Cold Ischemia time	CIT for kidneys transplanted in the program	Average value/minimum, average, maximum/distribution
Number of organs recovered from failed KEP matches	Organs initially recovered for a KEP matched transplant but used otherwise because of transplant cancellation	(total/average) over the reporting period
Outcome measures		
Number of rejection episodes	Number of rejection episodes among transplants within the KEP	Total/average number across the reporting period
Number of acute rejections	Number of acute rejections among transplants within the KEP	Total/average number across the reporting period
QALY for recipients	Quality-adjusted life years for recipients since transplant	Expected Total/average over multiple year horizon
QALY for donors	Quality-adjusted life years for donors since transplant	Expected total/average over multiple year horizon
Cost measures	Cost of transplantations including KEP maintenance costs	Total cost/cost per transplant across the reporting period

suggestion and approval. Measures of equity and fairness, e.g., in relation to PRA level, blood type, and ethnicity, have received little attention in the European literature and are not included.

The main and intuitive proxy for outcomes in KEPs has typically been the process indicator number of transplants, which is indeed the most reported measure. It signals that much of the reporting and assessment of KEPs has focused on the process rather than on the outcomes. This may be due to the need to focus on mastering and improving the operations of KEPs in the initial years of developing KEPs. It may alternatively be explained by the view that the relation between processes and outcomes are well-understood, and hence the outcome performance can be measured and managed by the process measures, such as the number of transplants. In any case, the process measures-subdivided into enrolment, matching, and transplantation measures-form the largest set of measures in KEPREPS and are pre-dominantly included in the Base Set.

The process and outcome measures require information on the context to be appreciated. Factors such as age, sensitization, blood type distribution, et cetera, are important to interpret process and outcome performance. Hence, the experts have considerably expanded the collection of measures providing context information, compared to the measures found by systematic literature review. Most of these measures are seen as important but not essential and are included in the Extended Set.

The Base set and Extended set of KEPREPS together facilitate unified scientific and practical reporting on KEPs.

Moreover, KEPREPS serves to enhance the practical relevance of model studies, which so far have differed quite substantially in their reporting, hampering their usefulness to inform and improve practice. In view of the importance presently attached to (health) outcomes and the difficulties faced globally to sustainably finance health systems, we believe that the inclusion of costs and outcomes in KEPREPS are especially valuable. Cost and health outcome measures have been hardly reported in scientific literature thus far. Hence the evidence base for the cost-effectiveness of KEPs is lacking, while a sound evidence base provides legitimacy to policy advancements of KEPS and especially larger coordinated KEPS which appear to be more effective than single center KEPS (23). Adoption of the KEPREPS standards by researchers and policy makers can therefore contribute to reducing the burden of ESRD while saving cost as the alternative of Dialysis is more expensive (38). More so, if KEPREPS may serve as a basis for reporting on model studies and the practice of emerging cross-national collaborations between KEPs (3) and future research expands it from a European standard to a global standard.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

BM: literature review, coordinating expert panel work, and writing. MM: writing and synthesis of results. JK: literature review and writing. All authors contributed to the article and approved the submitted version.

FUNDING

The authors acknowledge travel reimbursements by the COST program of the European Union (CA15210–European Network for Collaboration on Kidney Exchange Programmes). Article processing charges were funded by King Abdullah University of Science and Technology (KAUST).

ACKNOWLEDGMENTS

We are very thankful to the members of the expert panel: Peter Biro, Lisa Burnapp, Karine Hadaya, Antonio Nicolò, María Valentin, and five others. Furthermore, we thank the attendees of the ENCKEP Workshop in Glasgow. Finally, we thank Wichor Bramer for his help with the literature search.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.623966/full#supplementary-material

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Public Health

ORIGINAL RESEARCH
published: 12 August 2021
doi: 10.3389/fpubh.2021.600906

Efficacy Evaluation Study for Microburst Insulin Infusion: A Novel Model of Care

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Objectives: This study aims to evaluate the impact of Microburst Insulin Infusion (MII) treatment on Type 1 and 2 diabetic patients' HbA1c, lipids, peripheral neuropathy, and patient-reported health status.

Methods: We reviewed clinical charts, including lab results, for more than 80 diabetic and pre-diabetic patients treated at one U.S. outpatient clinic in St. Louis, Missouri between February 2017 and December 2019. Data included patient demographics, treatment data, lab and neuropathy tests, and self-reported patient health status questions.

The explanatory variable was number of months of MII treatment. Treatments are 3–4 h in length, with two intensive infusions the first week and one treatment each week thereafter, usually for 12 weeks total. Lab tests were at 12-week intervals.

Generalized linear modeling and t-tests assessed the significance of differences between patients' baseline lab values, neuropathy measures, and health status before treatment vs. after final treatment.

Results: Number of MII treatments per patient ranged from 1 to 262, over 1–24 months. Time in MII treatment was significantly associated with reductions in HbA1c by nearly 0.04 points per month, and triglycerides declined 3 points per month. Neuropathy measures of large toe vibratory sensation (clanging tuning fork) improved significantly, as did patient-reported health and feelings of improvement since beginning treatment.

Discussion: The MII therapy appears to be efficacious in treating diabetic patients, particularly those with complications like neuropathy. Our findings affirmed several other studies. We uniquely incorporated patient health questionnaires, and empirically studied MII treatment efficacy for diabetes in a population large enough to permit statistically valid inferences. With multiple waves of data for over 80 patients, this is one of the most extensive quantitative studies of microburst insulin infusion therapy conducted to date, with protocols more uniformly implemented and survey instruments more consistently administered by the same clinical team. Given the advances in insulin infusion therapy brought by MII, and early indications of its efficacy, the time is right for more in-depth studies of the outcomes patients can achieve, the physiological mechanisms by which they occur, MII's comparative effectiveness vis-à-vis traditional treatments, and cost-effectiveness.

Keywords: insulin infusion, efficacy, neuropathy, diabetes, diabetic complications

OPEN ACCESS

Edited by:
Stephanie Bernell, Oregon State University, United States

Reviewed by:
Connie J. Evashwick, George Washington University, United States
Vasco Fonseca, Universidade de Lisboa, Portugal

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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *31 August 2020*
Accepted: *05 July 2021*
Published: *12 August 2021*

Citation:
Howard SW, Zhang Z, Linomaz J, Lam W, Qian Z, Thurman J and BeLue R (2021) Efficacy Evaluation Study for Microburst Insulin Infusion: A Novel Model of Care. Front. Public Health 9:600906. doi: 10.3389/fpubh.2021.600906

INTRODUCTION

Diabetes mellitus is one of the most prevalent chronic diseases, affecting nearly 10% of the world's population, and imposing a cost of nearly $1 trillion USD globally (1–3). This chronic health condition is characterized by hyperglycemia which results from defects in glucose homeostasis stemming from relative or absolute deficiencies in insulin. Type 2 diabetes, comprising 90–95% of all diabetics, stems from a relative insulin deficiency as a result of the body becoming increasing resistant to the glucose lowering effects of insulin. Between 5 and 10% of diabetics are type 1, characterized by an autoimmune process with genetic predispositions and potential environmental factors that results in destruction of insulin-producing Beta cells in the pancreas (4). Complications from both Type 1 and Type 2 diabetes are an increasing public health challenge, and the prevalence of conditions like neuropathy can be as high as 50% among diabetics (5). Most research and clinical interventions have focused on managing blood sugar and/or insulin to healthy levels, which delays the onset of complications or slows their advance. Few interventions have been effective in improving the body's production or utilization of insulin. For patients in the outpatient environment, subcutaneous insulin injection is the most common management approach for insulin-resistant patients. In the inpatient environment, continuous insulin infusion may be used. While these interventions have helped slow the decline in the health of diabetics, they have not been able to halt or reverse complications such as neuropathy. In this exploratory study, we begin to evaluate the hypothesis that Microburst Insulin Infusion (MII) may be efficacious in treating HbA1c and complications of diabetes.

Limited research exists on the efficacy of MII, the pulsatile mode of insulin delivery most similar to that naturally occurring in the body. Earlier forms of insulin infusion included outpatient intravenous insulin therapy (OIVIT), pulsatile intravenous insulin therapy (PIVIT), hepatic activation therapy (HAT), chronic intermittent intravenous insulin infusion (CIIIT), metabolic activation therapy (MAT), and the Harvard Protocol. These techniques pulsed insulin via intravenous infusion, but at increasing levels that progressively built up insulin levels in the body. If the magnitude of these insulin pulses is reduced or absent there can be a defect in hepatic intracellular signaling resulting in increased hepatic output of glucose and reduced glucose utilization in animal models resulting in onset of Type 2 diabetes (6). In an animal study, researchers evaluated three different insulin delivery methods. One subgroup received pulsatile insulin (PII), one received a continuous insulin infusion while the last had reduced amplitude pulsatile insulin as seen in Type 2 diabetes. They found that those that received PII had improved insulin action, while those who received a stable insulin infusion or reduced amplitude pulses of insulin (as seen in Type 2 Diabetes) had abnormal liver metabolism resulting in increased liver production of glucose and worsening hyperglycemia with insulin resistance. Previous researchers further postulated the abnormal liver response may be contributing to some complications of diabetes such as abnormal lipid metabolism and cardiovascular disease (7). Other researchers have had similar findings in human studies, supporting the potential for MII to produce superior blood glucose management compared to continuous insulin infusion (8, 9).

MII is a newer mode of delivery and works differently. It aims to more closely simulate the function of a healthy pancreas, which delivers insulin from the pancreatic Beta cell to the circulation in a pulsatile fashion that varies with glucose levels in the bloodstream. MII aims to coordinate the timing and pulsed levels/quantities of insulin secretion, mimicking the workings of a healthy pancreas. These pulses are in approximate 5–6 min intervals and adjust insulin dosage to the glucose levels in the patient's blood (10, 11).

The literature includes early human studies of PII, in its various stages of evolution from PIVIT to MII. To mimic normal insulin delivery to the body, early researchers investigated patients with Type 1 diabetes mellitus on multiple daily injections of insulin and poor control. These patients were given 7–10 pulses of intravenous insulin over an hour while ingesting carbohydrates in the form of glucose. These sessions were given three times per day. After 2 days of treatments, therapy was then 1 day per week (12, 13). The literature contains more detailed explanation of the MII protocol and the mechanism by which it functions (10, 11). In a systematic review article by Dong et al. (10), the authors concluded that intravenous pulsatile insulin therapy in diabetics can lead to normal liver insulin levels as seen in non-diabetic individuals, and improvements in peripheral neuropathy symptoms (10). The liver in a Type 2 diabetic relies on metabolism of lipids over that of carbohydrates. It was also determined that pulsatile insulin therapy can change hepatic glucose metabolism favoring carbohydrate metabolism over lipid metabolism, thereby reducing free fatty acids which can promote inflammatory responses elsewhere (11). Given the findings of this research and the experiences of clinicians treating diabetics using MII, we purposed to study the efficacy of MII in controlling blood sugar, lipids, peripheral neuropathy, and patient-reported health status.

METHODS

Hypotheses and Methodological Strategy

We hypothesize that the longer a patient is treated with MII, the more their HbA1c, triglycerides, neuropathy, and self-reported health and wellbeing will improve, compared to the baseline of their first patient encounter. This is a retrospective cohort study delineating trajectories in each subject of laboratory and clinical measures from baseline. Should statistically significant improvement be observed for MII patients, a subsequent controlled trial may be in order.

Data

We reviewed clinical charts, including lab results, for all diabetic and pre-diabetic patients treated at one outpatient clinic in St. Louis, Missouri, USA between February 2017 and December 2019. There were 86 patients in total (**Table 3A**), of whom 60 had

lab data (of whom **Table 2** shows 52–56 had lab result data for each of the five outcome measures studied).

The explanatory variable of interest was the number of months (continuous) that patients were treated. The MII treatment sessions are ∼3–4 h in length. The patient receives two intensive intravenous infusions the first week of treatment (typically 1–2 days apart), and one treatment each week thereafter, usually for 12 weeks total. Some patients choose to continue receiving MII treatments longer term, though frequency may reduce to bi-weekly or monthly. Lab tests were drawn at ∼12-week intervals (a baseline panel of labs pre-treatment, another lab panel at week-12, and every 12 weeks thereafter for patients who opted to continue treatment).

The data abstracted from patient charts and questionnaires included lab values [HbA1c, total cholesterol, low-density lipids (LDL), triglycerides, and estimated glomerular filtration rate (eGFR)—all coded as continuous variables], patient demographics (age, sex, race, BMI, and diabetes type—all categorical variables), the duration of vibratory sensation from the CTF test (in seconds—continuous), and patient-reported health questions (0–10, 1–10, or percentage change—all coded as continuous). Each patient had between 1 and 262 MII treatments. At each treatment, a Family Nurse Practitioner (NP) administered the health questionnaire and clanging tuning-fork test (CTF) to assess neuropathy (14). The descriptive statistics are reported in **Table 3B**.

With the questionnaire, the NP asked each patient a series of standard self-reported health questions, coding their responses on the commonly-used 0–10 or 1–10 scale (i.e., "How is your diabetes-related pain on a scale of 1–10?"). The same Nurse Practitioner conducted all the patient questionnaires, and also recorded the duration of vibratory sensation from the big toe of each foot using a 128 Hz tuning fork (with a 15-s maximum/top-coding).

The questionnaire asked patients:

- How are you feeling today? (1–10 scale)
- How is your overall health? (1–10 scale)
- How is your diabetes-related level of pain? (1–10 scale)
- How does your diabetes-related pain interfere with your activities of daily living (ADLs)? (1–10 scale)
- How has your physical activity level changed since your last treatment? (% change)
- How has your energy level changed since your last treatment? (% change)
- How has your neuropathy changed since your last treatment? (% change)
- How has your sleep quality (including sleep pattern) changed since your last treatment? (% change)
- How has your vision changed since your last treatment? (% change)
- To what degree has your overall health changed since your FIRST treatment? (% change)

All the aforementioned measures were assessed at baseline and at each treatment session, and entered by the NP on the patient intake form. All data were subsequently abstracted into the analytic data set.

IRB Approval

The Institutional Review Board of the academic institution determined that ethical approval for this study was not required in accordance with local legislation and national guidelines, as no individually identifying information was available to the research team.

Statistical Modeling

Separate ordinary least squares regression models were run for the change in each outcome of interest (HbA1c, LDL cholesterol, triglycerides, total cholesterol, eGFR, vibratory sensation, and multiple self-reported health questions), comparing each patient's value at time of final (or most recent) treatment vs. the patient's baseline measures. Patient encounters that were missing data were omitted from the regression, and **Table 2** includes the number of patients with complete data included in each model (out of a total of 60 patients with lab results). Due to the smaller number of subjects, we used *t*-tests to gauge

TABLE 1A | Baseline descriptive statistics—lab data (categorical variables).

Variable	*N* (%) at baseline
Sex	
Male	24 (40.0%)
Female	36 (60.0%)
Race	
White	56 (93.3%)
Black/African–American	4 (6.7%)
Age (at first encounter)	
<45 years	9 (15.0%)
45–64 years	34 (56.7%)
≥65 years	17 (28.3%)
BMI category	
Normal or overweight	16 (26.7%)
Mild obesity	24 (40.0%)
Severe obesity	20 (33.3%)
Diabetes type	
Type 1	26 (43.3%)
Type 2	31 (51.7%)
Pre-diabetic	3 (5.0%)
HbA1c	
Well-controlled (<7.0)	16 (28.5%)
Fair-control (≥7.0, <9.0)	24 (42.9%)
Poor-control (≥9.0)	16 (28.5%)

TABLE 1B | Baseline descriptive statistics, continuous variables.

Variable	Mean	Minimum, Maximum
HbA1c	7.6	5.6, 10.9
eGFR	87.4	19, 173
LDL cholesterol	98.9	39, 170
Triglycerides	240.1	72, 617
Total cholesterol	170.4	106, 258

TABLE 2 | Regression models—lab data (changes in outcome measures).

Variable	HbA1c β (p-value)	LDL β (p-value)	Triglyceride β (p-value)	Total Cholest-erol β (p-value)	eGFR β (p-value)
Months in MII treatment	−0.038 (**$p = 0.067$**)	−0.291 ($p = 0.534$)	−3.115 (**$p = 0.035$**)	−0.440 ($p = 0.390$)	−0.045 ($p = 0.801$)
Male gender	−0.230 ($p = 0.558$)	−14.353 ($p = 0.079$)	62.776 (**$p = 0.013$**)	2.100 ($p = 0.809$)	−2.375 ($p = 0.441$)
Black/African American	−0.679 ($p = 0.397$)	−40.762 (**$p = 0.020$**)	9.659 ($p = 0.858$)	−42.617 (**$p = 0.029$**)	11.232 ($p = 0.098$)
Age (at first encounter)					
<45 years (vs. age 45–64)	0.061 ($p = 0.886$)	4.969 ($p = 0.611$)	−49.157 ($p = 0.102$)	3.038 ($p = 0.772$)	−6.401 ($p = 0.086$)
≥65 years (vs. age 45–64)	−0.129 ($p = 0.795$)	2.873 ($p = 0.790$)	−35.029 ($p = 0.308$)	2.957 ($p = 0.807$)	−5.552 ($p = 0.198$)
BMI category					
Mild obesity (vs. normal/overweight)	−0.460 ($p = 0.259$)	−7.07 ($p = 0.444$)	2.689 ($p = 0.925$)	−2.743 ($p = 0.787$)	−0.672 ($p = 0.853$)
Severe obesity (vs. normal/overweight)	−0.136 ($p = 0.757$)	−1.178 ($p = 0.906$)	14.180 ($p = 0.648$)	−2.323 ($p = 0.832$)	−6.349 ($p = 0.113$)
HbA1c control category					
Fair control (vs. good control)	−0.033 ($p = 0.939$)	N/A	N/A	N/A	N/A
Poor control (vs. good control)	−1.299 (**$p = 0.017$**)	N/A	N/A	N/A	N/A
Model R^2 goodness of fit statistic	0.371	0.188	0.238	0.143	0.236
Model F-statistic and p-value	3.02 (**$p = 0.007$**)	1.46 ($p = 0.208$)	2.10 ($p = 0.062$)	1.12 ($p = 0.368$)	1.99 ($p = 0.078$)
Number of patients w/usable data	56	52	55	55	53

Bold p-values indicate statistically significant coefficients.

the significance of differences between patients' continuous self-reported, subjective outcome measures before treatment vs. after final (or most recent) treatment. To test for non-linear relationships, we produced a scatter plot of HbA1c by weeks of MII treatment (**Supplementary Figure 1**). We also performed *t*-tests on the differences in self-reported, subjective measures between the baseline and the final treatments, and further stratified the statistics based on the categories of patients who only completed 1–5, 6–10, 11–15, or >15 treatments (**Table 4**). We used SAS University Edition version 2.8, to generate separate generalized linear models (GLM command) to identify statistically significant differences for each outcome variable between patients' first treatment and final treatment.

RESULTS: LAB TEST DATA

Descriptive statistics for the Lab Test analyses are presented in **Tables 1A,B**. Most patients were age 45–64 (56.7%), though 15% were younger than 45, and 28.3% older than 64. Sixty percent were female. All but 4 were white (93.3%). Three were prediabetic, 26 Type 1, and 31 Type 2 diabetics. With respect to weight, 26.7% were normal or overweight, 40% were mildly obese, and 33.3% were severely obese.

HbA1c measures ranged from 5.6 to 10.9 with a mean of 7.6 (**Table 1B**) (the CDC defines A1c levels below 5.7 as normal, 5.7–6.4 as pre-diabetic, and >6.4 as diabetic) 28.5% (16 patients) had well-controlled HbA1c; while 42.9% (24 patients) were moderately controlled ($7.0 \leq$ A1c < 9.0); and 28.5% (16 patients) had poorly controlled A1c ($\geq$9.0) (**Table 1A**). Estimated glomerular filtration rates (eGFR) ranged from 19 mL/min/1.73 m^2 to 173 mL/min/1.73 m^2 with a mean of 87.4 mL/min/1.73 m^2 (the CDC defines eGFR levels >90 ml/min as normal, 30–90 as mild or moderate reductions in kidney function, and <30 ml/min as severe reduction in kidney function) LDL ranged from 39 to 170 mg/dLwith a mean of 98.9 mg/dL (the CDC defines LDL levels <100 mg/dL as normal) Triglycerides ranged from 72 to 617 mg/dL with a mean of 240.1 mg/dL (the CDC defines Triglyceride levels <150 mg/dL as normal) Total cholesterol ranged from 106 to 258 mg/dL with a mean of 170.4 mg/dL (the CDC defines Total Cholesterol levels < 200 mg/dL as normal).

The results of each model for the Lab Test data are presented in **Table 2**. The primary explanatory variable of interest, the time variable for months of MII treatment, was associated with reductions in HbA1c levels by 0.038 A1c points per month, with a *p*-value of 0.067. This supported the hypothesis that MII can reduce triglycerides ($p = 0.035$), and HbA1c ($p = 0.067$) (Having poorly controlled HbA1c at baseline was also associated with reducing HbA1c). None of the other covariates had statistically significant associations in any of the models (Black/African–American race appears to be associated with lower LDL and Total Cholesterol levels, but there were only three African-Americans

in the data). The significance of the F-statistic and magnitude of R^2 coefficient indicate the model is a strong predictor of reduced HbA1c among the diabetic patients in the study.

RESULTS: NEUROPATHY AND PATIENT-REPORTED DATA

Separately, descriptive statistics for the Neuropathy and Patient-Reported Data Set (hereafter referred to as "Subjective Data") are presented in **Tables 3A,B**. Twenty-six patients had these data in their charts, but no lab results data. Consequently, the Subjective Data had 86 patients, whereas the lab data had only 60. Patient ages ranged from 19 to 85 with a mean of 57. Fifty-two were female and 34 were male. All but five were white. Four were prediabetic and averaged 11 treatments per patient during the study period. Fifty-three were Type 1 and averaged 28.6 treatments per patient, and 29 were Type 2 diabetics and averaged 24.5 treatments each. Number of MII treatments per patient ranged from 1 to 262.

Patients were asked to self-report on 10 health status questions, and the clinic NP measured vibratory sensation in both feet using a 128 Hz clanging tuning fork (CTF). Patients reported a mean of 9.1 and 9.4 s of vibratory sensation in the left and right feet, respectively, with a range of 0–15 s. Patients reported averages of 7.29 and 7.22 on the "How are you feeling?" and "How is your overall health?" questions, respectively, with ranges of 1–10. On the "How is your diabetes-related level of pain?" and "How is that pain interfering with your Activities of Daily Living (ADLs)?" questions, patients' average responses were 4.8 and 4.72, respectively, with ranges of 0–10. Patients were asked to rate their overall percentage health improvement since beginning treatment. Assessed at each treatment encounter, responses indicated a 47.2% improvement on average, ranging from −25% to 120%. The final five patient self-reported measures and their mean percentage changes were: physical activity change (33.4%), energy change (34.5%), neuropathy change (38.7%), changes in sleep patterns or sleep quality (39.4%), and vision change (24.2%).

The results of the *t*-tests are presented below in **Table 4**, including pre-/post-treatment differences and the *p*-values of their differences to indicate statistical significance ($p < 0.05$ indicates significant differences).

Statistically significant improvements of 3–3.5 s were observed for the CTF vibratory sensation for the right and left feet, respectively. Patient self-reports of feeling better and experiencing improved overall health, 0.54 and 0.69, respectively (on a scale of 1–10), were also statistically significant. Lastly, patients reported a statistically significant improvement, >25%, since beginning the MII treatment.

TABLE 3A | Descriptive statistics—self-report/subjective data (categorical variables).

Variable	*N* (%)
Sex	
Male	34 (39.5%)
Female	52 (60.5%)
Race	
White	81 (94.2%)
Black/African-American	5 (5.8%)
Age (at first encounter)	
<45 years	11 (12.8%)
45 to 64 years	47 (54.7%)
≥65 years	27 (31.4%)
BMI category	
Normal or overweight	16 (18.6%)
Mild obesity	24 (27.9%)
Severe obesity	19 (22.1%)
Diabetes type	
Type 1	53 (61.6%)
Type 2	29 (33.7%)
Pre-diabetic	4 (4.6%)
HbA1c	
Well-controlled (<7.0)	16 (18.6%)
Fair-control (≥7.0, <9.0)	22 (25.6%)
Poor-control (>9.0)	15 (17.4%)
(missing data)	33 (38.4%)

TABLE 3B | Descriptive statistics—self-report/subjective data (continuous variables).

Variable	Mean	Minimum, Maximum
Age (years)	57	19, 85
Total # of MII treatments per patient	24.7	1, 262
How are you feeling?	7.29	1, 10
How is your overall health?	7.22	1, 10
How is your diabetes-related pain?	4.80	1, 10
Diabetes pain interference w/ ADLs?	4.72	1, 10
Vibratory sensation—right foot (seconds)	9.37 s	0, 15
Vibratory sensation—left foot (seconds)	9.13 s	0, 15
Overall improvement since starting MII?	47.2%	−25, 120%
Physical activity change? (since last treatment)	33.4%	−65, 200%
Energy change? (since last treatment)	34.5%	−65, 100%
Neuropathy change? (since last treatment)	38.7%	−40, 100%
Sleep pattern/quality change? (since last treatment)	39.4%	−50, 100%
Vision change? (since last treatment)	24.2%	−10, 100%

DISCUSSION

We aimed to study relationships between patients' number of weeks in MII treatment (changes between baseline and last or most recent treatment) and their associated HbA1c, triglycerides, neuropathy symptoms, and self-reported health. Overall, patients experienced improvements in both lab values and self-reported measures the longer they were in treatment. **Table 4** showed short-term reductions in self-reported health scores, and the NP and clinic medical director posited that patients' reports of how they are feeling, diabetic-related pain and overall health may worsen in the short-run as neuropathy symptoms diminish,

TABLE 4 | *T*-tests: significance of pre-/post-differences in outcome measures—patient-reported/subjective data.

Variable	Overall pre-/post: last visit vs. baseline	Categorized: 1–5 visits vs. baseline	Categorized: 6-10 visits vs. baseline	Categorized: 11–15 visits vs. baseline	Categorized: 16–30 visits vs. baseline
Vibratory sensation—right foot (seconds)	2.99 (**$p < 0.001$**)	2.67	1.86	1.27	4.32
Vibratory sensation—left foot (seconds)	3.46 (**$p < 0.001$**)	3.67	1.29	2.43	4.00
How are you feeling?	0.54 (**$p = 0.011$**)	−0.45	1.18	0.50	0.79
How is your overall health?	0.69 (**$p < 0.001$**)	−0.34	0.18	0.67	1.29
How is your diabetes-related pain?	−0.52 ($p = 0.131$)	0.00	−0.85	0.33	−0.33
Diabetes pain interference w/ADLs?	0.01 ($p = 0.977$)	−1.00	−0.70	0.57	−1.05
Overall improvement since starting MII?*	25.38% (**$p < 0.001$**)	7.17%	6.15%	23.91%	38.88%
Physical activity change? (since last treatment)	NS	NS	NS	NS	NS
Energy change? (since last treatment)	NS	NS	NS	NS	NS
Neuropathy change? (since last treatment)	NS	NS	NS	NS	NS
Sleep pattern/quality change? (since last treatment)	NS	NS	NS	NS	NS
Vision change? (since last treatment)	NS	NS	NS	NS	NS

**MII, Microburst Insulin Infusion therapy. Bold p-values indicate statistically significant coefficients.*

sensation returns, and they begin to experience pain again. Our generalized linear models show HbA1c declining approximately 0.038 A1c points per month, and triglyceride levels declining ∼3 mg/dl per month in MII therapy. This finding is in line with, and potentially higher than the ∼4–12% reductions observed over 1–4 years in other studies, including results from studies of metformin or metformin plus additional drugs (15), sulphonylureas (16), TZD drugs (17), and inhaled insulin (18). Vibratory sensation measures also improved, as did patients' self-reported measures of how they were feeling, and their overall health also improved at statistically significant levels. These findings are also in line with results published over the past 10–20 years, including those of Elliott et al. (11) (8–13, 19–23).

Previous Research

Our findings were consistent with some of the earlier research on PII generally, or MII more specifically. Multiple studies supported the hypothesis that pulsed insulin infusions could be more efficacious than continuous insulin infusion (8, 9, 20–22). Our results were also consistent with the findings of the systematic literature review by Dong et al. (10) and Elliott et al. (11), we also found improvements in neuropathy (10, 11). However, most studies of MII have focused on metabolic measures estimated from patients' respiratory O_2 and CO_2 (10, 11). We focused on the lipid, HbA1c, eGFR, neuropathy measures, and patient self-reported health screening questions more commonly used in ambulatory care clinics globally. Our study is differentiated by incorporating the differences between MII and earlier pulsatile insulin infusion approaches, longer period of study (up to 3 years), a relatively large sample size, stronger methodology, and focus on clinical measures more commonly used by clinicians treating diabetics. Taken together, these differentiating factors make the current research an important addition to the body of literature on diabetes care.

Strengths

This is one of the few studies to empirically study the efficacy of the MII treatment for diabetes in a population large enough to permit statistically valid inferences. With multiple waves of data on over 80 patients, this is one of the most extensive quantitative studies of microburst insulin infusion therapy conducted to date, with protocols more uniformly implemented and survey instruments more consistently administered by the same clinical team. Consistent with our hypotheses, we found statistically significant improvements in HbA1c, triglycerides, neuropathy, and 3 self-reported patient survey measures of health and well-being, supporting the hypothesis that the MII therapy is efficacious in treating diabetic patients, particularly those with complications like neuropathy. This study is also one of the few to implement surveys of patients' self-reported health and quality of life. Like the findings of Dong et al. (10) and Elliott et al. (11), patients reported significantly improved health after the 12-week course of treatment, and frequently even longer-term.

Limitations

While this study is pioneering, it has inherent limitations. Our hypothesis was that MII would show early indications of potential efficacy, using solely existing chart data for patients served from February 2017 through December 2019. Given frequent contact with patients over time, the Hawthorne effect and regression to the mean may have impacted results. In the current retrospective cohort study context, we were not able to include a control group,

and the *t*-tests and regression models effectively used subjects as their own controls over weeks of time in treatment. Sample size is also a concern. While this is one of the largest studies yet conducted on the MII treatment, statistical power was reduced, limiting our options for statistical modeling. This may also have contributed to lack of statistical significance of the findings for low-density lipids (LDL cholesterol) and estimated glomerular filtration rate (eGFR), measures we expected to improve with time in treatment.

The CDC estimates over 30 million Americans have diabetes, and the WHO estimated the global count at over 400 million in 2014 and identify the disease as a major cause of heart disease, stroke, blindness, and amputations (2, 3, 24, 25). In 2017, diabetes-related costs of care were ∼$10,000 per diabetic patient, with the disease imposing billions of dollars in indirect costs just in the U.S. alone (25). Better managing the degenerative effects of diabetes would not only decrease pain and suffering of patients, but could also save trillions of dollars in direct and indirect costs worldwide.

Given the promising preliminary findings of this retrospective study, further research is warranted. The research team plans a second phase of the study to compare MII results observed to-date with results observed among a retrospective cohort of diabetic patients managed through traditional lifestyle modification (diet and exercise) treatment protocols at their affiliated academic medical center. If that study shows comparatively superior outcomes, a multi-center, prospective clinical trial should be pursued. Given the advances in insulin infusion therapy brought by MII, and early indications of its efficacy, the time is right for more in-depth studies of the outcomes patients can achieve, the physiological mechanisms by which they occur, MII's comparative effectiveness vis-à-vis traditional treatments, and cost-effectiveness.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available, subject to approval by the clinic leadership.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Saint Louis University Institutional Review Board. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements. Data were provided de-identified to the researchers, and the study was deemed exempt.

AUTHOR CONTRIBUTIONS

SH and ZZ managed the entirety of the study. JL and WL provided valuable assistance with literature review, data entry, and writing. ZQ, JT, and RB provided important subject matter consultation, advice on study design, and contributed to the writing process. All authors contributed to the article and approved the submitted version.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.600906/full#supplementary-material

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Conflict of Interest: SH previously consulted on a clinical improvement and strategic planning project with Mitokon Health Center, the St. Louis clinic from which the de-identified data were obtained.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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frontiers in Public Health

ORIGINAL RESEARCH
published: 24 August 2021
doi: 10.3389/fpubh.2021.589734

Consumers' Knowledge, Attitudes, and Practices Toward Medicine Price Transparency at Private Healthcare Setting in Malaysia

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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: 31 July 2020
Accepted: 02 August 2021
Published: 24 August 2021

Citation:
Ahmad NS, Hatah E, Jalil MR and Makmor-Bakry M (2021) Consumers' Knowledge, Attitudes, and Practices Toward Medicine Price Transparency at Private Healthcare Setting in Malaysia. Front. Public Health 9:589734. doi: 10.3389/fpubh.2021.589734

Background: Medicine price transparency refers to the practice of making prices available to consumers for them to identify, compare, and select the medicine that provides the desired value. This study aimed to evaluate consumer knowledge, attitudes, and practices regarding Malaysia's medicine price transparency initiative, as well as factors that may influence related good consumer practices in private healthcare settings.

Methods: A cross-sectional, self-administered survey was conducted between May and July 2019 among consumers attending private healthcare institutions in Malaysia. The self-developed and validated survey consisted of four sections on the following: respondents' demographics, and 28 close-ended and graded Likert scale answer options on knowledge, attitudes, and practices toward medicine price transparency. Factors influencing good consumer practices toward the transparency initiative were modeled using binary logistic regression.

Results: A total of 679 respondents were part of the study. The mean age of respondents was 38 ± 13.3, with the majority (n = 420, 61.9%) being female. The respondents' mean score of knowledge and attitudes toward the price transparency initiative was 5.6 ± 1.5 of the total score of 8 and 31.9 ± 4.0 of the total score of 40, respectively. The respondents had the lowest score in the practice of price transparency, with a mean score of 31.5 ± 5.6 of the total score of 60. Male gender, Chinese ethnicity, high score on knowledge and attitudes, and high expenses on medicines influenced respondents' good practices of medicine price transparency.

Conclusion: Respondents had good knowledge and attitudes, but their usage and implementation of the medicine price transparency initiative was still inadequate. A number of factors influence this inadequacy, including gender, race, consumers' out-of-pocket spending on medication, and knowledge of and attitudes toward price transparency practices. Consumer-driven market price control would be impossible to achieve without the good consumer practices of medicine price transparency.

Keywords: price transparency, medicine price, consumer, knowledge, attitude, practice

INTRODUCTION

Price transparency in pharmaceuticals have been used in several countries as a strategy to help reduce expenditure on medicines. It can be defined as the practice of making prices available to consumers and/or to the government or the authority responsible for controlling or setting the market price of medicines. This is usually achieved through various mechanisms, such as the prices being published on the government/relevant website, displayed at the healthcare facilities, and printed on medicine labels or consumers' receipts and medical bills (1, 2). Australia, New Zealand, Lebanon, Oman, and Tunisia are examples of countries known to participate in medicine price transparency (3). They have accomplished this by publishing the prices on their government's website for the use of consumers. This transparency initiative helps consumers and the government to identify, compare, and choose medicines that offer the desired value (4). It creates awareness of price discrimination, which leads to informed choices and cost-saving by the users (5–9). It is likely to save the consumers' out-of-pocket spending by helping them with value-based purchasing and allowing them to exercise their right to price information before purchase (10, 11). Nonetheless, communication, education, and information about good medicine purchasing behavior are reported to have an impact on overall consumer behavior (12). Thawani et al. discovered that in India, after an information, education, and communication intervention, consumers' awareness of drug price variation, attitudes toward expensive and brand medicines, and behavior of comparing drug price information improved among 500 consumers (12). They concluded that consumers use medicines based on their knowledge, perceptions, and habits.

In Malaysia, the healthcare system is divided between public and private. The public healthcare system is offered to all Malaysian citizens at a low cost as it is highly subsidized by the government (13). The private healthcare system is funded through private insurance, employer benefits, and out-of-pocket payment. Although the public healthcare system is highly subsidized, patients may also choose to seek treatment in private healthcare settings such as retail pharmacies, private hospitals, and clinics (13). The government controls the price of medicines in the public healthcare system through direct negotiation and bulk purchasing. Nonetheless, in the free market, there is no price control for medicines supplied in private healthcare settings (14). It is estimated that 60% of pharmaceutical usage in private healthcare comes from consumers' out-of-pocket expenses (15).

Due to the lack of price control in private healthcare, the medicine prices are reported to be high and varied (14, 16–18). Based on a Ministry of Health (MOH) study over 2011–2015, the markup for generic and innovator medicine prices was between 31–402% (median 96%) and 24–86% (median 39%), respectively (14). The Malaysian government implemented the medicine price transparency initiative in 2011 as part of the National Medicine Policy (MNMP) to ensure consumer access to affordable medicines (19). The initiative includes the pharmaceutical industry's voluntary disclosure of medicine reference prices to the government and the public (20). Since the policy's launch in 2011, Malaysian consumers' knowledge, attitudes, and practices regarding medicine price transparency have remained unstudied. Although the National Survey on the Use of Medicines (NSUM) discovered that 68% of consumers believe that price label information is helpful in making an informed decision when purchasing medicines, it is not known whether this is actually practiced in real life (21). Other government initiatives aimed at increasing price transparency include strengthening the provision of itemized billing, which specifies the price of each item at all dispensing outlets, thus allowing for price comparison and reporting by the public (19). Despite the government's initiative to encourage consumers to practice medicine price transparency when purchasing medicine or receiving treatment in the private healthcare system, it is unclear whether Malaysian consumers have used the provision of itemized billing.

Therefore, the purpose of this study was to assess consumers' knowledge, attitudes, and practices regarding medicine price transparency initiatives, such as itemized billing and price comparison, as well as to investigate factors that may influence consumers good medicine purchase practices in Malaysia's private healthcare settings. The study also sought to shed light on whether it is possible to control medicine market prices in private healthcare through consumers' good purchasing practices.

MATERIALS AND METHODS

Study Design and Sampling

This study was conducted as a cross-sectional survey among the public between May and July 2019. Individuals aged 18 years and above, Malaysian citizens, and those with experience in out-of-pocket purchases from private healthcare facilities in Malaysia were invited to participate in the study. Using the Raosoft sample size calculator for a survey study and a confidence interval of 95%, a margin of error of 5%, and a Malaysian adult population of 22 million (22), an estimated sample size of 385 was required. Respondents were excluded from the study if they did not complete 80% of the questionnaire, understand English or Malay, or provide informed consent. Using convenient sampling, the self-administrated survey was distributed face-to-face and online using a Google form shared through social media channels, such as Facebook and WhatsApp. The face-to-face survey was distributed across the country in public places in urban and rural areas, such as shopping malls, community pharmacies, clinics, and community halls. Participation in the study was entirely voluntary and without remuneration. Respondents who agreed to participate were asked to sign the informed consent form or click the agreement button before answering the survey questions.

Survey Instrument

The questionnaire was developed through a literature review of reports and documents related to the medicine price transparency initiative (21, 23, 24) and inputs from domain experts. It consisted of four sections: (a) respondents' demographics and characteristics, (b) knowledge—8 items, (c) attitudes—8 items, and (d) practices related to consumer rights on medicine purchasing—12 items. Section A gathered respondents' information on age, gender (male or female),

race (Malay, Chinese, Indian, or other races), highest level of education, occupation, monthly income, area of residence (urban or rural), medical coverage (insurance health policy or employer medical coverage), healthcare condition, amount of money spent on medicine, facilities where medications were usually obtained, and method used to access medicine price information. In the remaining sections, respondents were asked about their knowledge, attitudes, and practices regarding their rights when receiving medications, such as price information, itemized billing, and filing a complaint if any problem. They were provided with "yes," "no," or "not sure" answer options in Section B, five Likert-scale answer options ranging from "strongly agree" to "strongly disagree" in Section C, and "never" to "always" in Section D. The score for each section was calculated using the sum of the scores for a correct answer or a score between one to five for the statement with negative attitudes or "never practice," and most positive attitudes and "always" practice. Reverse-scoring was given for all negative statements accordingly. Using Bloom's cut-off point, respondents' practice score on medicine price transparency was categorized as good if they had a sum score of 60% and above, which is a combination of a high and moderate score, and poor for a score <60% (25).

Data Analysis

The content validity of the questionnaires was evaluated by two academicians, two MOH pharmacy officers, and two independents reviewers who were experts in medicine pricing and/or consumer surveys. The content validity index (CVI) and the average scale-level CVI (S-CVI/Ave) was conducted to measure the relevancy and clarity of the statements and its proportion relevance judged by all expert (26). The result of the item-CVI (I-CVI) for statements in each section was between 0.83 and 1, with S-CVI/Ave of 0.93, 0.96, and 0.92 for the knowledge, attitudes, and practices sections, respectively. The survey was initially prepared in English and translated to Malay by two independent translators using backward and forward translation. To ensure clarity and reliability, the questionnaires were pilot tested with 30 members of the public. The Cronbach's alpha coefficient (r) for Sections B, C, and D were found to be reliable at 0.70, 0.76, and 0.75, respectively (27).

The data were analyzed using the IBM SPSS software Version 24 for descriptive and inferential analyses. Factors likely to influence respondents' good practices of rights when purchasing medicines, with a score of >60%, were modeled using binary logistic regression with a stepwise-backward approach. The variables tested were respondents' demographics and characteristics, as well as their knowledge and attitudes score on medicine purchasing behavior, with a $p < 0.05$ considered significant. Prior to the binary logistic regression, a univariate analysis was performed to determine which variables would be included in the final model analysis. Variables were included if the adjusted odds ratio had $p < 0.25$ (28).

RESULTS

A total of 679 responses were received for the study, with 406 completed face-to-face and 273 completed online. Ten respondents were excluded from the study because their responses were <80% complete. As a result, a final 679 responses were included in the analysis. A summary of the respondents' demographics and characteristics is presented in **Table 1**. The mean age ± standard deviation of the respondents was 38 ± 13.3, with a majority ($n = 420$, 61.9%) being female and of Malay ethnicity ($n = 459$, 67.6%). A total of 422 (65.1%) had a diploma or bachelor's degree as their highest education level. The majority ($n = 465$, 68.5%) worked and had a monthly income of less than RM3000 ($n = 382$, 56.2%). More than half ($n = 421$, 60.0%) had private insurance or employer benefit coverage and were healthy. Only 135 respondents (19.9%) had serious health problems, such as cardiovascular disease, diabetes mellitus, hypercholesterolemia, asthma, gastroenteritis, and arthritis. A total of 353 (52.0%) respondents spent less than RM100 in a month for medicine, while a majority ($n = 414$, 61.0%) reported checking medicine price information before purchasing a medicine, with most practicing comparing the printed price at various healthcare facilities ($n = 338$, 49.8%).

The respondents' mean score of knowledge of the medicine price transparency initiative was 5.6 ± 1.5 from the maximum score of 8. A majority of the respondents knew they had the

TABLE 1 | Respondents' demographics and characteristics.

Variables	Descriptions	*N* (%)/Mean (s.d)
Gender	Male	259 (38.1)
	Female	420 (61.9)
Age		38 (13)
Race	Malay	459 (67.6)
	Chinese	120 (17.7)
	Indian	62 (9.1)
	Others	38 (5.6)
Highest education level	College/University	422 (65.1)
	Upper middle school	153 (22.5)
	Lower school	15 (2.2)
Occupation	Employed	465 (68.5)
	Unemployed/housewife	66 (9.7)
	Student	122 (18.0)
	Pensioner	26 (3.8)
Monthly income (RM)	<1,000	206 (30.3)
	1,000–3,000	176 (25.9)
	3,001–6,000	181 (26.7)
	>6,000	116 (17.1)
Living area	Urban	565 (83.2)
	Rural	114 (16.8)
Private insurance health coverage	No coverage	258 (38.0)
	Private insurance/Employer benefit	421 (62.0)
Health status	Has health problem	135 (19.9)
	Do not has heath problem	544 (80.1)
Medicine expenditure per year (RM)	<100	353 (52.0)
	100–500	256 (37.7)
	500–1,000	44 (6.5)
	>1,000	26 (3.8)

right to get their medication anywhere they preferred ($n = 517$ 76.1%), were aware that the price of the same medicine could be different at different healthcare facilities ($n = 550$, 81.0%), and that they were entitled to receive an itemized bill from their health practitioners ($n = 629$, 92.5%). However, only 318 (46.8%) of the respondents knew about the medicine price guide on the Pharmaceutical Service Programme (PSP) website. A total of 258 (38.0%) were not sure they could make a complaint to the MOH about overcharging on medical and medicine bills. **Table 2** presents a summary of the respondents' scores on knowledge of the medicine price transparency initiative in Malaysia.

The mean score on respondents' attitudes toward medicine price transparency in Malaysia was 31.9 ± 4.0 of the total score of 40. A majority of them ($n = 539$, 79.4%) "strongly agreed" and "agreed" about the need and importance of being aware of the medicine cost before purchase for making an informed choice ($n = 598$, 88.1%). A total of 604 respondents (88.9%) "strongly agreed" or "agreed" that price comparison helped them get the best price for their medicines. There were 571 (84%) respondents who "disagreed" with the need for requesting itemized bills, yet the majority ($n = 623$, 92.8%) "strongly agreed" and "agreed" that an itemized bill should include the price of each medicine rather than the total cost. A total of 211 (31.1%) respondents were "neutral" about paying high prices for medicines in private healthcare settings, while 155 (22.8%) "agreed" or "strongly agreed" that they did not mind paying higher prices for medicines in a private clinic or hospital. **Table 3** presents a summary of the attitudes score of the respondents.

Next, the respondents' mean score on practices related to medicine price transparency was 31.5 ± 5.6 of the total score of 60. A total of 125 respondents (18.4%) "never" asked the estimated price of medicines before receiving treatment, while 420 (61.81%) "always" and "very often" complied with the medicine choice made by their doctor, regardless of price. Only a small number of respondents ($n = 108$, 15.9%) "always" asked for an itemized bill. More than half "never" negotiated nor asked for a price discount when purchasing medicines at a retail pharmacy ($n = 376$, 55.4%) and private health clinics and hospitals ($n = 471$, 69.4%). Only 64 (9.4%) respondents "always" asked their healthcare provider for an explanation of speculative charges on their bill. **Table 4** presents a summary of respondents' practices score on the price transparency initiative.

The respondents' practices scores were then classified as good or poor using a percentage score that is the sum of the scores divided by the total score multiplied by 100. Only 220 (32.4%) had good practices, with a score of ≥60%, and the rest ($n = 459$, 67.6%) had poor practices in medicine price transparency, with a score of <60%. Gender, race, annual spending on medicines, knowledge, and attitudes scores were found to have a significant influence on respondents' good practices in price transparency [$\chi^2_{(11,N=679)} = 75.56$, $p < 0.001$]. Male respondents were 1.78 times more likely than female respondents to apply good practices in medicine price transparency (AOR [95% CI] = 1.78 [1.26, 2.56], $p < 0.001$). Chinese (AOR [95% CI] = 1.96 [1.26, 3.04], $p = 0.003$) and other races (AOR [95% CI] = 2.29 [1.12, 4.67] $p = 0.023$) were also more likely to apply good practices than the Malays. The study also discovered that respondents who spent more than RM1000 on their medicines were 3.13 times more likely to practice good medicine price transparency than those who spent less than RM100 per month (AOR [95% CI] = 3.13 [1.33, 7.36], $p = 0.009$). Furthermore, a 1% increase in the knowledge and attitudes scores increased the likelihood of respondents engaging in good practices by 41% (AOR [95% CI] = 1.41 [1.24, 1.61], $p < 0.001$) and 6% (AOR [95% CI] = 1.06 [1.04, 1.11], $p < 0.001$), respectively. The final model on factors that influence respondents' good practices on medicine price transparency is presented in **Table 5**.

DISCUSSION

This study provides an overview of Malaysian consumers' knowledge, attitudes, and practices regarding medicine price transparency in private healthcare settings. It found that the majority of respondents scored higher in knowledge of and attitudes toward the medicine price transparency initiative than in practice. One related area where respondents were found to have a lack of knowledge concerns the medicine price guide on the PSP website (29). This could be because the website was relatively less known to the public, and/or respondents preferred to physically check prices at the facilities rather than online. Therefore, increasing medicine price transparency physically in the healthcare settings, as practiced in stores or clinics in the Philippines and hospitals in Thailand, will be a good alternative to increasing consumers' medicine price transparency practices (1, 2). Since 2019, private hospitals in Thailand have been required to display their medicine prices on their advertisement board, website, or via QR scan codes (2). If such a practice is implemented in Malaysia, consumers would be able to verify the medicine prices using scanned QR codes from the healthcare facilities of their choice (2). Checking drug prices before purchasing them would protect consumers from being overcharged and provide them with an opportunity to discuss their concerns about drug costs with their doctors (30). Furthermore, it can increase consumer confidence in negotiating drug pricing, allowing them to obtain their medication at reasonable prices and continue their treatment affordably. More information, education, and communication interventions are also required to improve Malaysian consumers' behavior when purchasing medicines or receiving treatment in private healthcare settings.

In this study, a majority of the respondents (91.8%) agreed on the importance of obtaining itemized bills following their treatment. Nevertheless, this did not translate into practice as only 15.9% of the respondents "always" practiced obtaining itemized bills. This could be because, first, the Malaysian Private Healthcare Facilities and Services Regulation specifies that itemized bills are required in private healthcare settings only if the patient requests for it (31, 32). As a result, providing itemized bills for patients is not a usual practice, particularly in primary healthcare settings such as clinics and pharmacies. Second, because itemized billing is a non-voluntary practice,

TABLE 2 | Respondents' knowledge of medicine price transparency in private healthcare setting in Malaysia.

No	Statement	Yes *N* (%)	No *N* (%)	Not sure *N* (%)
1.	I have the right to get my medication anywhere at my preference, not necessarily from the doctor who is treating me.	517 (76.1)	103(15.2)	59 (8.7)
2.	I can request a change for a cheaper medication from my doctor/ pharmacist.	366 (53.9)	169 (24.9)	144 (21.2)
3.	Every patient is entitled to get a itemized bill from health practitioners (doctors or pharmacists).	629 (92.5)	11 (1.6)	39 (5.7)
4.	It is sufficient if the itemized bill lists only the total costs of the treatment and medications.	195 (28.7)	368 (54.2)	116 (17.1)
5.	I have the right to know the estimated charges for my treatment.	566 (83.4)	37 (5.4)	76 (11.2)
6.	I can check the medication price guide at the Pharmaceutical Service Programme, Ministry of Health website.	318 (46.8)	42 (6.2)	319 (47.0)
7.	The price of the medication at private hospitals, clinics and retail pharmacies may differ regardless of the same brand.	550 (81.0)	22 (3.2)	107 (15.8)
8.	Complaints about overcharging medical and medicine bill can be lodged with the Ministry of Health Malaysia.	398 (58.6)	23 (3.4)	258 (38.0)

TABLE 3 | Respondents' attitudes on medicine price transparency practice in private healthcare setting in Malaysia.

No	Statement	Strongly agree *N* (%)	Agree *N* (%)	Neutral *N* (%)	Disagree *N* (%)	Strongly disagree *N* (%)
1.	A patient should know the costs of treatment beforehand.	273 (40.2)	266 (39.2)	101 (14.9)	27 (4.0)	12 (1.8)
2.	Price comparison before purchasing helps consumers to get the best price for their medication.	337 (49.6)	267 (39.3)	57 (8.4)	10 (1.5)	8 (1.2)
3.	I need not be concerned about the medication price if it is covered by my insurance company or my employer.	41 (6.0)	111 (16.3)	129 (19)	250 (36.8)	148 (21.8)
4.	I do not mind paying high price of medicine in a private clinic or hospital.	32 (4.7)	123 (18.1)	211 (31.1)	194 (28.6)	119 (17.5)
5.	It is not fair to set a high medicine price to those with higher income.	203 (29.9)	216 (31.8)	153 (22.5)	73 (10.8)	34 (5.0)
6.	The itemized bill should list out the price of each medication prescribed rather than just the total cost in a bill.	351 (51.7)	272 (40.1)	45 (6.6)	5 (0.7)	6 (0.9)
7.	It is not important to ask for itemized billing.	13 (1.9)	29 (4.3)	66 (9.7)	276 (40.6)	295 (43.4)
8.	It is important to know the medication charge to allow me to make a better choice.	292 (43)	306 (45.1)	65 (9.6)	10 (1.5)	6 (0.9)

patients may perceive receiving a non-itemized bill as good consumer behavior and would not ask for an itemized bill. Finally, experience with compliance with non-itemized bills may be a reason why patients do not request them. According to a study on private hospital billing in Malaysia, some hospitals did not follow the suggestion for itemized billing, for example, when the treatment costs such as for medicines were presented as a lumpsum amount or in combination with other item costs such as consumables (33). To ensure compliance with the itemized billing regulation, stricter penalties and monitoring are necessary. In addition, instead of issuing itemized bills only on patient request, the government may consider making them mandatory, leading to a better selection of medicines by patients and prevention of overcharging.

The respondents were also found to be more likely to comply with a doctor's choice of medicine, regardless of price, and less likely to negotiate on the medicine price or ask for a discount. This could be because consumers are more concerned about receiving effective treatment regardless of cost, as reported in previous studies (30, 34), or because they really trust their healthcare providers (35). The study by Schafheutle et al. in England reported that a majority of their patients rarely and reluctantly discussed medicine prices and their affordability with their general practitioners (30). This is often because they felt reluctant to discuss due to the short consultation time, or, as stated, did not want to jeopardize the relationship they had with their doctor. Similarly, Fraeyman et al.'s study of patients with chronic diseases in Belgium revealed that <4% of the participants discussed medicine price issues with their doctors or pharmacists (36). Thus, for healthcare providers to include and initiate discussions about medicine prices and affordability with their patients should be encouraged.

TABLE 4 | Respondents' practice on medicine price transparency practice in private healthcare setting in Malaysia.

No	Statement	Always *N* (%)	Very often *N* (%)	Sometimes *N* (%)	Rarely *N* (%)	Never *N* (%)
1.	I ask the estimated price of medication before getting any treatment.	104 (15.3)	122 (18.0)	204 (30.0)	124 (18.3)	125 (18.4)
2.	I check for the price of medications before purchasing at retail pharmacy.	144 (21.2)	200 (29.5)	145 (21.4)	101 (14.9)	89 (13.1)
3.	I compare the prices at different pharmacies prior deciding to purchase the medications.	97 (14.3)	148 (21.8)	183 (27)	136 (20)	115 (16.9)
4.	I ask for a cheaper brand of medication when I cannot afford the recommended medication price.	55 (8.1)	127 (18.7)	167 (24.6)	145 (21.4)	185 (27.2)
5.	I comply with the doctor's choice of medication regardless of the price.	134 (19.7)	286 (42.1)	172 (25.3)	60 (8.8)	27 (4.0)
6.	I ask for the itemized bill every time I seek treatment from any private clinic or hospital.	108 (15.9)	156 (23)	135 (19.9)	133 (19.6)	147 (21.6)
7.	Besides the price, I take into consideration the quality and effectiveness when purchasing medications.	261 (38.4)	256 (37.7)	86 (12.7)	40 (5.9)	36 (5.3)
8.	I ask for discount/negotiate the price of my medication at retail pharmacy.	32 (4.7)	45 (6.6)	82 (12.1)	144 (21.2)	376 (55.4)
9.	I ask for discount/negotiate the price of my medication at private clinic or hospital.	15 (2.2)	26 (3.8)	40 (5.9)	127 (18.7)	471 (69.4)
10.	I pay for my medication even if I feel that the price unreasonable.	96 (14.1)	229 (33.7)	186 (27.4)	71 (10.5)	97 (14.3)
11.	I ask for an explanation from my healthcare provider regarding any speculative charge in my bill.	64 (9.4)	172 (25.3)	140 (20.6)	115 (16.9)	188 (27.7)
12.	I complain to the authority if I feel that I am billed unreasonably.	15 (2.2)	52 (7.7)	52 (7.7)	97 (14.3)	463 (68.2)

In total, 28% of the respondents of this study stated that they "rarely" or "never check" their medicine price before purchasing. This is similar to the 2013 study by Baber and Ibrahim on consumer attitudes on affordability of medicines in Malaysia, where 37% of the respondents did not check the medicine price before purchasing them (23). This demonstrates that, even after many years, consumer behavior in purchasing medicine has not changed and must be urgently improved for their welfare.

In this study, male gender, Chinese ethnicity, high knowledge and attitudes scores, and high medicine expenditure cost were found to influence good consumer practices regarding medicine price transparency. In line with previous NSUM findings, all demographic variables, including race and ethnicity, were significantly associated with medicine price label checking and purchasing behavior (21). Because knowledge and attitudes can influence good consumer behavior in terms of medicine price transparency, it is critical to educate and communicate with consumers on a regular basis to raise their awareness and practice of price transparency. Interventions such as printed handouts, press releases, and interactive discussion sessions on medicine pricing have been found to be effective in changing consumer behavior in India, such as comparing prices before purchasing and being concerned about aspects of medicine use (12). Furthermore, respondents who had a high out-of-pocket expenditure on medicines were more likely to use the price transparency initiative to reduce their treatment costs (37).

There are some limitations to this study. First, patients who can afford to pay for the service or who have health insurance or employer benefit coverage are more likely to use private healthcare services in Malaysia. As a result, this study may have excluded people who could not afford treatment in private healthcare settings. Second, because socioeconomic status influences treatment choice in Malaysia, with the wealthier seeking care in private healthcare settings (38), the study's findings may be influenced by the differences in consumer socioeconomic status and payment schemes. Nonetheless, respondents from various socioeconomic backgrounds were included in this study to ensure that the findings were generalizable to the public on average. Third, the cross-sectional study design represents the findings at one point in time and does not reflect future findings of consumers' knowledge, attitudes, and practices on medicine price transparency. Last, the nature of the survey required patients to recollect their purchasing behavior practices, which may be open to recall bias, which is common to such survey study designs (39).

CONCLUSION

In summary, despite good knowledge and attitudes scores among the consumers, the practice of attaining medicine price transparency is still unsatisfactory and inadequate in Malaysia. A number of influencing factors were found, including gender, race, consumers' out-of-pocket spending on medicines, and knowledge and attitudes scores in price transparency practices. Consumer-driven market price control would be impossible to achieve without good consumer practices related to price

TABLE 5 | Factors that may influence respondents' good practice toward medicine price transparency at the private healthcare setting.

Variables	Poor practice *n* = 459 *n* (%)[a]	Good practice *n* = 220 *n* (%)[a]	Univariate analysis (*n* = 679)			Multivariate analysis (*n* = 679)		
			Crude OR (95% CI)	Wald's χ^2 (*df*)	*P*-value	Adj. OR (95% CI)	Wald's χ^2 (*df*)	*P*-value
Gender								
Male	157 (60.6)	102 (39.4)	1.00	9.24 (1)	0.020	1.00	10.48 (1)	<0.001
Female	302 (71.9)	118 (28.1)	0.60 (0.43, 0.83)			0.56 (0.39,0.79)		
Race								
Malay	329(71.7)	130 (28.3)	1.00	11.77 (3)	0.008	1.00	12.44 (3)	0.006*
Chinese	68 (56.7)	52 (43.3)	1.94 (1.28, 2.93)		0.002	1.96 (1.26, 3.04)		0.003*
Indian	40 (64.5)	22 (35.5)	1.39 (0.79, 2.43)		0.246	1.22 (0.67, 2.22)		0.510
Others	22 (57.9)	16 (42.1)	1.84 (0.94, 3.62)		0.077	2.29 (1.12, 4.67)		0.023*
Highest education								NS
College/University	349 (68.3)	162 (31.7)	1.00	1.53 (2)	0.466			
Upper middle school	102 (66.7)	51 (33.3)	1.08 (0.73, 1.58)		0.705			
Lower school	8 (53.3)	7 (46.7)	1.89 (0.67, 5.29)		0.228			
Occupation								NS
Employed	311 (66.9)	154 (33.1)	1.00	7.30 (3)	0.063			
Unemployed/housewife	49 (74.2)	17 (25.8)	0.70 (0.39, 1.26)		0.233			
Student	87 (71.3)	35 (28.7)	0.81 (0.53, 1.26)		0.352			
Pensioner	12 (46.2)	14 (53.8)	2.36 (1.06, 5.22)		0.035			
Monthly income (RM)								NS
No income and <1,000	143 (69.4)	63 (30.6)	1.00	1.34 (4)	0.855			
1,000–3,000	119 (67.8)	57 (32.4)	0.92 (0.53, 1.62)		0.778			
3,001–6,000	121 (66.9)	60 (33.1)	0.96 (0.55,1.67)		0.872			
>6,000	76 (65.5)	40 (34.5)	1.01 (0.56, 1.85)		0.965			
Location of living								
Urban	382 (67.6)	183 (32.4)	1.00	0.00 (1)	0.989			NS
Rural	77 (67.5)	37 (32.5)	1.0 (0.65, 1.54)					
Health coverage								NS
No coverage	185 (71.7)	73 (28.3)	1.00	3.19 (1)	0.074			
Private Insurances/Employer benefit	274 (65.1)	147 (34.9)	1.36 (0.97,1.90)		<0.001			
Disease status								NS
Has health problem	88 (65.2)	47 (34.8)	1.00	0.45 (1)	0.503			
No health problem	371 (68.2)	173 (31.8)	0.87 (0.57, 1.30)					
Medicine expenditure per year (RM)								
<100	251(71.1)	102 (28.9)	1.00	12.37 (3)	0.006	1.00	7.89 (3)	0.048*
100–500	172 (67.2)	84 (32.8)	1.20 (0.85, 1.70)		0.300	1.17 (0.81, 1.69)		0.398
500–1,000	26 (59.1)	18 (40.9)	1.70 (0.89, 3.24)		0.105	1.56 (0.79, 3.07)		0.201
>1,000	10 (38.5)	16 (61.5)	3.94 (1.73, 8.97)		0.001	3.13 (1.33, 7.36)		0.009*
Age	37.5[b] (12.5)[c]	39.9[b] (14.4)[c]	1.02 (1.00, 1.03)	5.42 (1)	0.020			NS
Knowledge score	5.4[b] (1.5)[c]	6.1[b] (1.3)[c]	1.46 (1.29, 1.66)	34.92 (1)	<0.001	1.41 (1.24, 1.61)	26.51 (1)	<0.001
Attitude score	31.6[b] (4.1)[c]	32.7[b] (3.5)[c]	1.08 (1.03, 1.13)	12.17 (1)	<0.001	1.06 (1.01, 1.11)	6.52 (1)	<0.011

**Significant p < 0.05.*

[a]The percentage is reported by column, adding up to 100% based on available information.

[b]Mean.

[c]SD.

COR, crude odds ratio; AOR, adjusted odd ratio; CI, confidence interval; NS, non-significant.

transparency, such as asking for itemized bills, and checking, comparing, and negotiating the price of medicines. Aside from educating and raise consumer awareness about the importance of medicine price transparency, government intervention such as compulsory itemized bills and increase medicine price transparency physically in the healthcare settings are required.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Human Research Ethics Committee, Universiti Kebangsaan Malaysia (UKM PPI/111/8/JEP-2019-060). The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

NA conceptualized and designed, conducted data collection, analyses, and drafted the initial manuscript. EH and MM-B conceptualized and designed the study, interpretation of data and reviewed, and revised the manuscript. MJ conceptualized, conducted data collection, and revised the manuscript. All contributors approved the final manuscript as submitted and had complete access to the study data that support the publication.

FUNDING

This study received the Fundamental Research Grant Scheme (FRGS) from the Ministry of Education, Malaysia (FRGS/1/2018/SS03/UKM/03/2).

ACKNOWLEDGMENTS

We would like to thank the experts who have helped to validate our survey and respondents who have participated in the study. We also thank the Director General of Health Malaysia for his permission to publish this article.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Public Health

REVIEW
published: 27 October 2021
doi: 10.3389/fpubh.2021.750122

The Linkages Between Reimbursement and Prevention: A Mixed-Methods Approach

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Background: The benefits of prevention are widely recognized; ranging from avoiding disease onset to substantially reducing disease burden, which is especially relevant considering the increasing prevalence of chronic diseases. However, its delivery has encountered numerous obstacles in healthcare. While healthcare professionals play an important role in stimulating prevention, their behaviors can be influenced by incentives related to reimbursement schemes.

Purpose: The purpose of this research is to obtain a detailed description and explanation of how reimbursement schemes specifically impact primary, secondary, tertiary, and quaternary prevention.

Methods: Our study takes a mixed-methods approach. Based on a rapid review of the literature, we include and assess 27 studies. Moreover, we conducted semi-structured interviews with eight Dutch healthcare professionals and two representatives of insurance companies, to obtain a deeper understanding of healthcare professionals' behaviors in response to incentives.

Results: Nor fee-for-service (FFS) nor salary can be unambiguously linked to higher or lower provision of preventive services. However, results suggest that FFS's widely reported incentive to increase production might work in favor of preventive services such as immunizations but provide less incentives for chronic disease management. Salary's incentive toward prevention will be (partially) determined by provider-organization's characteristics and reimbursement. Pay-for-performance (P4P) is not always necessarily translated into better health outcomes, effective prevention, or adequate chronic disease management. P4P is considered disruptive by professionals and our results expose how it can lead professionals to resort to (over)medicalization in order to achieve targets. Relatively new forms of reimbursement such as population-based payment may incentivize professionals to adapt the delivery of care to facilitate the delivery of some forms of prevention.

Conclusion: There is not one reimbursement scheme that will stimulate all levels of prevention. Certain types of reimbursement work well for certain types of preventive care services. A volume incentive could be beneficial for prevention activities that are

OPEN ACCESS

Edited by:
Steven W. Howard,
Saint Louis University, United States

Reviewed by:
Jason Scott Turner,
Rush University Medical Center, United States
France Weaver,
Xavier University, United States

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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *30 July 2021*
Accepted: *29 September 2021*
Published: *27 October 2021*

Citation:
Zwaagstra Salvado E, van Elten HJ and van Raaij EM (2021) The Linkages Between Reimbursement and Prevention: A Mixed-Methods Approach. Front. Public Health 9:750122. doi: 10.3389/fpubh.2021.750122

easy to specify. Population-based capitation can help promote preventive activities that require efforts that are not incentivized under other reimbursements, for instance activities that are not easily specified, such as providing education on lifestyle factors related to a patient's (chronic) disease.

Keywords: prevention, reimbursement, incentives, primary prevention, secondary prevention, tertiary prevention, quaternary prevention, rapid review methods

INTRODUCTION

Healthcare prevention, ranging from regular dental cleaning to collective initiatives to promote a healthier lifestyle, is one of the most important pillars of public health (1). Major gains in health can be accomplished through prevention (2). Moreover, prevention has the potential to substantially reduce disease's economic burden (3), especially in the current environment of growing chronic illness (4). Healthcare prevention focuses on promoting and protecting people's health by ensuring they receive care that conforms to their needs and stage of disease (5). While primary, secondary, and tertiary prevention focus on delivering care to avoid disease onset, allow early diagnose and reduce disease impact, respectively (6), quaternary prevention aims to protect patients from receiving redundant, unnecessary care (7). Healthcare professionals face the challenge of having to promptly assess a patient's need for preventive interventions (8, 9).

This crucial deliberation could, however, be disrupted by incentives in reimbursement systems (10). In healthcare systems with a purchaser-provider split, third-party funders such as insurers or the government can pay professionals for the services provided based on different types of reimbursement schemes (11). Reimbursement schemes can vary on many aspects, such as unit of payment (e.g., per service, per patient, or per day), payment amount, or timing (11). Different combinations of these characteristics are argued to create different (financial) incentives that promote or hinder professionals' behavior in everyday practice, e.g., providing less or more services than necessary (10, 11). As for pay-for-performance (P4P), one of its reported perverse incentives is that it might focus providers' attention to what is being measured, and consequently marginalize other quality criteria that are not being rewarded (12).

All-in-all, reimbursements have a well-documented reputation for incentivizing unwanted behavior. However, in the same train of thought, well-designed reimbursement schemes may allow the possibility to incentivize the behavior we do want, i.e., to focus professionals on prevention. Therefore, reimbursement schemes may play an important role in supporting meaningful prevention (13).

Much research has been devoted to investigating how various reimbursement schemes (and their respective incentives) impact healthcare delivery (11, 14–16). However, the existing body of literature still lacks comprehensive research on the impact of reimbursement schemes on professionals' behavior on all four levels of prevention. This study addresses this gap by incorporating evidence from both a rapid review and original empirical research, to address our research question: How do different types of reimbursement schemes in healthcare affect healthcare professionals' behavior in terms of the delivery of prevention?

METHODS

To address our research question, we use a mixed-methods approach (17). We conduct both a rapid literature review for a broad overview of the literature and semi-structured interviews with healthcare professionals for more in-depth insights. In this section, we present the research methods.

Rapid Review

We review the literature using a rapid review methodology. With more widely established systematic reviews, time and resource consumption may pose as barriers for its use in strategic decision making and health policy formulation. Rapid reviews are known for providing information on a specific research topic within a limited timeframe, applying systematic review methodology with explicitly stated shortcuts whilst maintaining rigorous methodology (18). The shortcuts applied to tailor our rapid review are specifically stated as the use of one database and one main reviewer. As progress toward universal health coverage should be informed by timely evidence, rapid reviews are an efficient approach to producing relevant evidence often to support decision-makers and strengthen health policies (19).

For our review, studies were systematically identified using the online database PubMed. Only English language scientific articles were considered for which full text was available, published from 2010 up until April 25th 2020, using the Pubmed "Humans" filter. Different search strings were tested using five previously identified relevant papers. To ensure that the relevant studies were included, the final search string was achieved based on the results of this testing. The search string can be found in the **Supplementary Material**.

The inclusion and exclusion criteria were defined *a priori*. All types of academic primary research studies (empirical studies and conceptual works) were considered. Editorials, systematic reviews, and studies reporting and/or commenting on data from other studies were excluded. The population of interest is healthcare professionals, broadly defined as qualified medical professionals who deliver primary, secondary, and/or tertiary healthcare services. Therefore, the search string contains various terms that are used to describe different types of healthcare professionals. As previously described, it is expected that reimbursement schemes and payment models induce a variety of behaviors in professionals, thus impacting the delivery of prevention. The phenomenon of interest comprises reimbursement schemes' effect on primary, secondary, tertiary

and/or quaternary prevention. Professionals' behaviors expressed in process and/or outcome measures were included as long as it pertained to primary, secondary, tertiary and/or quaternary prevention. Studies that do not define the specific type of reimbursement under analysis were excluded, as well as studies that analyze the effect of reimbursement on prevention in combination with other interventions without isolating the effect of reimbursement. For example, a study from Kalwij et al. (20) examines the impact of financial incentives combined with practice-based support (audits and feedback) on performance and on screening behavior, instead of the isolated effect of financial incentives. This led to its exclusion during full text screening.

Our rapid review's search string yielded 3,591 papers from PubMed. **Figure 1** illustrates the inclusion and exclusion process. One author (ES) screened titles and abstracts. This resulted in 75 papers eligible for full-text screening. Full-text screening led to the exclusion of 48 studies due to reasons such as type of reimbursement is not specified, or link to prevention is not clear. Each step in this process was discussed within the author team, before as well as during execution of each step. A total of 27 conceptual or empirical papers published between January 2010 and April 2020 were included for the qualitative synthesis, all related to the effect of reimbursement of healthcare professionals on delivery of prevention.

Two authors (HE and ER) provided feedback and assisted in calibrating eligibility criteria, screening and selecting papers, and cross-checking data extraction. The relevant data from all included studies were extracted and collected in an Excel spreadsheet. These results were posteriorly synthesized according to the type of reimbursement and prevention level(s) addressed and analyzed in terms of the relationships between reimbursement types and preventive behaviors.

Semi-structured Interviews

We conducted semi-structured interviews to further our understanding of the subject matter. We chose this qualitative method to collect in-depth data and capture meanings and perceptions people attribute to a certain phenomenon (22). In conjunction with the rapid nature of our research and limited by COVID-19 restrictions, our number of interview participants is limited. In total, ten semi-structured in-depth interviews were conducted with eight Dutch healthcare professionals—consisting of four general practitioners (GP) and four physical therapists (PT)—as well as two members of the prevention and purchasing departments of Dutch insurance companies (representing the payers in the Dutch healthcare system). An overview of respondents' characteristics can be found in the **Supplementary Material**.

In the Netherlands, GP practices are reimbursed through a 3-segment funding model. In the first segment, GPs are reimbursed through a mix of capitation and FFS for primary care activities provided. The second segment consists of funding through episode-based payments where GPs receive a fixed fee for every patient for which they provide multidisciplinary care such as diabetes care and other selected chronic diseases. In segment 3, GPs and insurance companies have the opportunity to negotiate additional P4P contracts and GPs become eligible to receive a bonus for reaching certain outcomes (at practice level) pertaining to the care delivered in segments 1 and 2.

Physical therapy in the Netherlands is reimbursed under FFS, in other words, paid according to the number of physical therapy sessions. Practices are free to make specific agreements with purchasers regarding delivery of care, volume or outcomes in exchange for financial rewards. Professionals under different types of reimbursement were purposely selected and answers were analyzed according to the type of reimbursement.

All semi-structured interviews were held between March and April 2020 and were conducted via telephone due to the COVID-19 outbreak. Interviews were recorded and transcribed verbatim in Dutch and the quotes used in this manuscript were posteriorly translated to English. Interviews were coded and analyzed using software ATLAS.ti, version 8.4.4. The interview transcripts were analyzed first using open coding and subsequently using axial coding to integrate codes into categories and identify relationships between categories. Examples of codes include "Obstacles for prevention," "Efforts made toward prevention," "Perceptions about own role in prevention" and "Strategies to mitigate overmedicalization." The code group list can be found in the **Supplementary Material**.

As previously mentioned, qualitative research allows for the collection of in-depth data such as perspectives and perceptions; elements that would be much more difficult to obtain from quantitative data (22). However, these essential elements that provide an extra dimension and enrich findings of qualitative research are also the subject of controversies regarding quality and trustworthiness of its results. Therefore, besides the reliance on multiple research methods, we have incorporated other strategies in this research to enhance its validity and reliability. Concerning the empirical part of this research, interviews were recorded to ensure descriptive validity and increase reliability. The use of in-depth open interviews helps mitigate interpretation bias and consequently increase internal validity. Interviews were transcribed *verbatim* and transcripts were made available to increase internal reliability. The use of a topic list to guide interviews helped mitigate researcher bias regarding assumptions or beliefs that might otherwise have compromised validity. The topic list can be found in the **Appendix**. The interviewing process as well as data collection, analysis and interpretation steps were discussed between the authors and are described in detail allowing for a well-documented audit-trail of materials and processes. Regarding our rapid review, we tested different search strings in order to find the search string that yields as many relevant studies and thus achieve higher sensitivity.

RESULTS

In this section, the research findings are presented. We present the rapid review's findings organized by type of reimbursement scheme and complement these with relevant findings from the semi-structured interviews.

The rapid review yielded 27 studies; their respective characteristics are presented in more detail in **Table 1**. Another

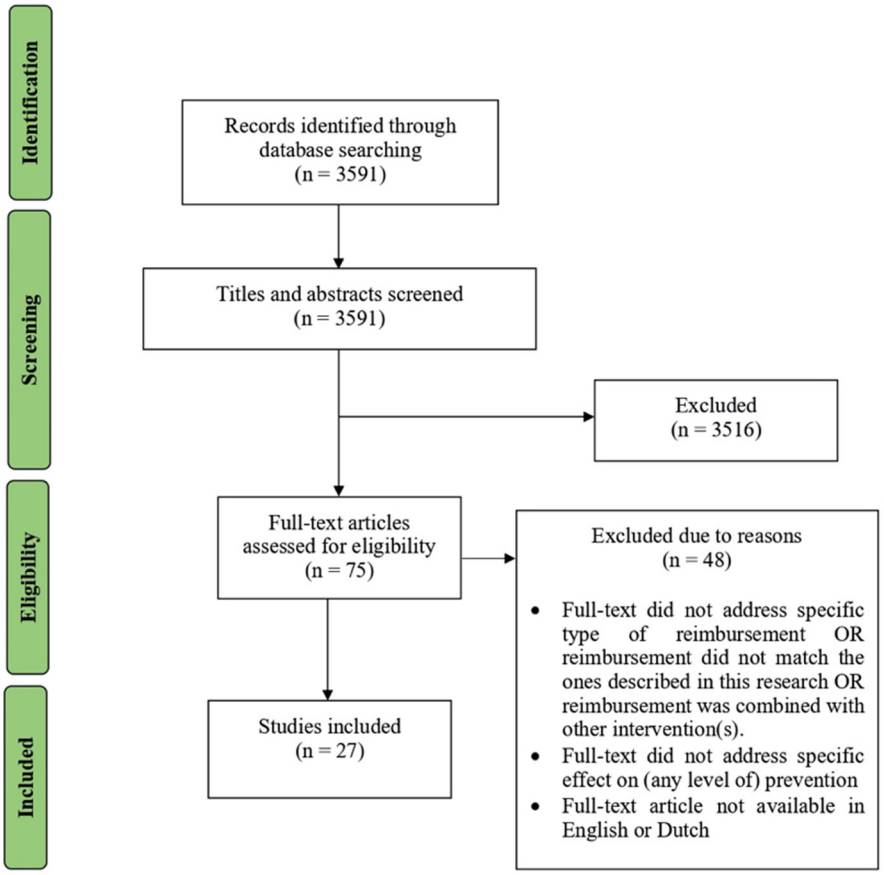

FIGURE 1 | Flow diagram of inclusion and exclusion process based on PRISMA flow diagram (21).

table with more detailed information about the included studies can be found in the **Supplementary Material**. The reviewed studies were executed in a variety of countries and delivery models. More than half (14 out of 27) in a country with a National Health Insurance (NHI) model: eight in Canada (27, 29, 35, 36, 38, 40, 41, 46), five in Taiwan (24, 25, 31, 37, 44), and one in Rwanda (28). A further five in a country with a Beveridge model: four from the UK (34, 39, 43, 48) and one from Italy (32). Four studies were executed in the US, with their focus varying from publicly funded safety-net community health centers (30), Medicaid-focused managed care (26), a commercial health plan (23) to a cross-sector study (45). Two studies in a country with a Bismarck model: one in Estonia (42) and one in France (49). One study was executed in Mozambique and concerned a donor-sponsored program (47). Finally, one study (33) analyzed data from 14 different European countries, including countries with a Bismarck model (such as The Netherlands) and countries with a Beveridge model (such as Sweden).

As presented in **Table 1**, a total of 20 studies focus on the relationship between P4P bonuses and prevention (23, 24, 26, 28, 30–32, 34, 35, 37–40, 42–44, 46–49). Seven papers study P4P incentives awarded at practice level (26, 28, 34, 39, 43, 47, 48), from which four studies pertain to the Quality and Outcome Framework (QOF) P4P program in the UK, where incentives represent up to 25% of annual income (34, 39, 43, 48). In one study, an extra bonus is awarded to practices that, besides achieving the targets, also manage to do this within a short period of time (26).

The remaining 13 studies on P4P consider bonuses directed at individual professionals (23, 24, 30–32, 35, 37, 38, 40, 42, 44, 46, 49), from which four studies specify that incentives represent between 2 and 4% of professional's income (23, 30, 35, 42) and 1 $<$ 10% (40). Two studies (31, 44) consider a program where an additional bonus (on top of P4P for achieving targets) is awarded to professionals who rank in the top 25%.

From all 20 studies considering P4P, eight studies do not further specify bonus characteristics besides bonus amount and if directed at practice or at individual level (24, 28, 32, 37, 38, 46, 47, 49).

From the remaining seven studies that do not address P4P (25, 27, 29, 33, 36, 41, 45), four studies compare the impact of multiple payment models (FFS vs. salary vs. capitation) (27, 33, 41), or different blends of FFS, capitation and incentives (36), on professionals' behavior toward prevention. One study

TABLE 1 | Overview of included studies in rapid review.

Reference	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
Chen et al. (23)	Longitudinal retrospective study	USA Commercial Health Plan (not government-run)	Secondary Tertiary	P4P Bonus (3.5% above reimbursement: maximum $16,000/year). Performance based on physician level	To assess the impact of the P4P program on improved quality of care (lipid monitoring and treatment) and quality of care on outcomes (new coronary events, hospitalizations, and lipid control) for cardiovascular disease.	Primary care setting and medical specialized care	P4P was associated with a higher likelihood of receiving quality care (OR = 0.70; 95% CI = 0.54–0.90) compared to non-P4P. Receiving quality care was then associated with a lower likelihood of new coronary events (OR = 0.80; 95% CI = 0.69–0.92), hospitalization (OR = 0.76; 95% CI = 0.69–0.83), or uncontrolled lipids (OR = 0.67; 95% CI = 0.61–0.73), $p < 0.01$.
Chen et al. (24)	Controlled before and after study	Taiwan NHI model	Secondary Tertiary	P4P Bonus for completion of visits $3/visit/patient + additional bonus for further screening, referral, early detection of abnormalities per patient ($15–$30). Performance based on physician level	To evaluate the effect of a P4P program targeting providers' performance on three indicators for Hepatitis B and C guideline-recommended preventive services.	Hospital and clinic physicians	P4P was associated with a significantly higher likelihood of receiving all three recommended services (OR = 1.13; 95% CI: 1.07–1.19). The before and after difference between the two groups was modest (5–23%).
Cheng et al. (25)	Population-based natural experiment	Taiwan NHI model	Secondary Tertiary	Episode-based vs. FFS	To examine the impacts of diagnosis-related group (DRG) payments on health care provider's behavior (medical service content and healthcare outcomes) compared to the traditional FFS.	Hospital setting	DRG payment resulted in a decrease of 10% ($p < 0.001$) in length of stay in the intervention group in relation to the comparison group. The number of orders that define intensity of care declined significantly ($p < 0.001$) with differences of 1.230, 2.695, and 1.070 items. No significant changes were found at $p < 0.001$ significance level for healthcare outcomes variables.
Chien et al. (26)	Case-comparison and interrupted times series	USA Not-for-profit Medicaid-focused managed care plan	Primary	P4P Piece-rate bonus (15–25% above reimbursement) Bonus for immunizations $100/patient and extra bonus $100/patient for timeliness. Performance based on practice level	To evaluate the impact of a "piece-rate" P4P program on fully and up-to-date immunization of 2-year-olds.	All types of healthcare practices (not further specified)	Results on the fourth year of intervention show that the P4P healthcare program presented significantly (OR = 0.60, SE = 0.12, $p < 0.01$) higher fully and timely immunization rates than the comparison group.

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
Dahrouge et al. (27)	Cross-Sectional study	Canada NHI model	Primary Secondary	FFS vs. Salary vs. New capitation (Capitation + 10% FFS) vs. Traditional capitation	To compare delivery of preventive services (immunizations and screenings) by practices under four different primary care funding models and to identify organizational factors associated with superior preventive care.	Primary care (family health networks; health services organizations; community health centers)	After adding physician characteristics and organizational structure factors in multilevel regression analysis, reimbursement was (no longer) statistically significant. Having at least one female family physician ($\beta = 8.0$, 95% CI 4.2–11.8), a panel size of fewer than 1,600 patients per FTE family physician ($\beta = 6.8$, 95% CI 3.1–10.6) and an electronic reminder system ($\beta = 4.6$, 95% CI 0.4–8.7) were more significant.
De Walque et al. (28)	Prospective quasi-experimental study	Rwanda NHI model	Primary secondary	P4P Bonus (different amounts for different services: individual testing US$0.92; couple testing US$4.59). Performance based on practice level	To examine the impact of a P4P incentive on two output indicators: individual and couples HIV testing and counseling.	Healthcare Facilities (not further specified)	The impact of P4P on HIV testing and counseling was significant for individuals living in couple (β (estimated effect) = 0.102, SE = 0.041, $p = 0.012$) and for discordant couples ($\beta = 0.147$, SE = 0.068 $p = 0.0130$) and not significant for individuals not living in couple ($\beta = 0.003$, SE = 0.062, $p = 0.959$) and not discordant couples ($\beta = 0.072$, SE = 0.070, $p = 0.304$).
Échevin and Fortin (29)	Natural experiment	Canada NHI model	Secondary Tertiary	Per-diem	To examine the impact of hospital specialists' reimbursement on length of stay (LOS) and re-hospitalization post-discharge as an alternative to traditional FFS.	Specialist physicians at hospital setting	Under the new per-diem reimbursement there was an increase in the LOS by 0.28 days (SE = 0.07) corresponding to 4.2% increase. The reform did not impact the risk of re-hospitalization at a global level ($\beta = -12{,}798$, $p > 0.202$).
Gavagan et al. (30)	Retrospective analysis of a natural quasi-experiment	USA Community Health Program (CHP) Clinics	Primary Secondary	P4P Bonus [max $12,000 annually/physician ($4,000/target = 3–4% annual income)]. Performance based on physician level	To evaluate physician P4P program on quality of preventive care (childhood immunization and cervical- and breast cancer screenings).	Primary care (community health centers)	P4P was associated with slight improvements in performance for mammography ($p = 0.076$) and cervical cancer screenings ($p = 0.053$, however this was not considered clinically significant. The effect on immunizations was not significant ($p = 0.79$). Survey results point out that physicians felt the incentives were not very effective in improving quality of care.

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
Hsieh et al. (31)	Longitudinal cohort study	Taiwan NHI model	Secondary Tertiary	P4P Diabetes program. Phase 1–process indicators–Bonus for process indicators ($30.00–$75.00/visit); Phase 2–Bonus ($30.00/visit) for process indicators conditional on performance of outcome indicators. Ranking (top 25% performing physician get extra bonus). Performance based on physician level	To examine if a change in P4P (from a program with process measures to process and outcome measures) had impact on diabetes outcomes.	Medical specialized care (hospitals and clinics)	The provision of tests for HbA1c [0.001, 95% CI = (0.000–0.003) p = 0.154] and LDL [0.019, 95% CI = (−0.017–0.055) p = 0.302] did not significantly differ between both phases. Blood pressure examinations significantly increased [0.068, 95% CI = (0.032–0.103) $p < 0.001$] between phases. Adding outcome measures in the second phase led to significant improvement in HbA1c [−3.135 95% CI = (−3.818–2.453) $p < 0.001$] and LDL levels [−4.323 95% CI = (−6.004–2.643) $p < 0.001$].
Iezzi et al. (32)	Longitudinal cohort study	Italy Beveridge Model	Secondary Tertiary	P4P "low powered incentives". Performance based on physician level	To analyze the impact of a low powered P4P incentive on diabetes management. The outcome measure is set on the number avoidable hospitalizations.	Primary care (general practitioners)	The results (available upon request) associated financial incentives from P4P with a lower likelihood of experiencing avoidable hospitalizations for diabetes-related diseases.
Jusot et al. (33)	Cross-sectional study	14 European countries Various health delivery models	Primary Secondary	FFS vs. capitation vs. salary	To examine the variations in utilization of preventive services (immunization and screenings) in 14 European countries. One of the health system supply determinants being remuneration methods for physicians.	Entire healthcare system	FFS was associated with a higher probability for colon cancer screening (OR = 3.038 significant at 1%) compared to capitation (OR = 0.593) or salary (OR = 0.395 significant at 1%). Similar results were presented for flu vaccinations. The results associate capitation with the lowest provision of eye exams (OR = 0.493 significant at 1%) while FFS was associated with the highest score (OR = 2.084 significant at 1%).
Karunaratne et al. (34)	Prospective longitudinal cohort study	UK Beveridge model	Tertiary	P4P Quality and Outcomes Framework (QOF) (representing 25% of income). Performance based on practice level	To evaluate the effectiveness of adding renal indicators to P4P program on hypertension management in primary care patients with chronic kidney disease (CKD) by analyzing changes in recorded blood pressure and prescription patterns before and after their introduction.	Primary care (general practices)	In general, blood pressure (BP) reduced between period 1 and 2 and was sustained in period 3. There was a more pronounced effect in the hypertensive patients (both CKD and not) as mean BP went from 146/79 mmHg to 140/76 in the first 2 years post-P4P ($p < 0.01$) and was sustained in the last 2 years of the study [139/75 ($p < 0.01$)]. Within the hypertensive group the CKD patients had a BP greater reduction, the % of patients with BP reduced to 145/85 mmHg went from 28 to 45.1 to

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
							55.6%. The BP reduction was associated with an increase in medication prescription and consequently increased prescription costs from €444.726 to €655.842).
Kiran et al. (35)	Longitudinal study	Canada NHI model	Secondary	P4P Bonus (max $8.400/annually—3% of gross income) for reaching screening targets. Performance based on physician level	To assess whether the introduction of a P4P reimbursement scheme was associated with increased cancer screening rates and also its effect on physician payments.	Primary care—PCMH (patient-centered medical homes)	No significant change was found for screening rates after introduction of P4P. E.g., Colon cancer screening rate changed from 3.0% (95% CI, 2.3–3.7%) to 4.7% (95% CI, 3.7–5.7%). Financial incentives for cervical, breast, and colorectal cancer screening accounted for $28.3, $31.3, and $50.0 million expenses respectively between 2006 and 2010.
Kiran et al. (36)	Longitudinal study	Canada NHI model	Secondary Tertiary	Different combinations of FFS and capitation	To understand the effect of each payment model on chronic disease management and prevention (cancer screenings) over time, comparing the effectiveness of different models.	Primary care—PCMH (patient-centered medical homes)	Compared to enhanced fee-for-service, team-based capi-tation was associated with a higher likelihood of performing dia-betes monitoring (39.7 vs. 31.6%, adjusted RR = 1.22, 95% CI = 1.18–1.25), mammography (76.6 vs. 71.5%, adjusted RR = 1.06, 95% CI = 1.06–1.07) and colorectal cancer screening (63.0 vs. 60.9%, adjusted RR = 1.03, 95% CI = 1.02–1.04). Over time, absolute difference in improvement in diabetes monitoring of team-based capitation compared with enhanced fee for ser-vice [10.6% [95% CI 7.9–13.2%)] and with non–team-based capitation [6.4% (95% CI 3.8–9.1%)]. Absolute difference in improvement in cervical cancer screening of team-based capitation com-pared with enhanced fee for service [7.0% (95% CI 5.5–8.5%)] and compared with non-team-based capitation [5.3% (95% CI 3.8–6.8%)]. No significant differ-ences over time for breast and colorectal cancer screening rates.

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
Lai and Hou (37)	Cross-Sectional study	Taiwan NHI model	Secondary Tertiary	P4P Diabetes Program. Physicians receive fees for enrolling patients in program + Incentives for process indicators and Outcome indicators. Performance based on physician level	To examine the effect of a diabetes mellitus P4P program (DM-P4P) on guideline adherence for diabetes mellitus disease management according to physician participation status. Patients were divided in three groups: patients enrolled in the DM-P4P program, patients not enrolled but treated by DM-P4P-participating physicians, and patients treated by non-P4P physicians.	Physicians (hospitals and practices)	DM-P4P program was associated with a higher likelihood of receiving all 7 guideline-recommended tests/examinations ($p < 0.001$). Patients who were not enrolled in the program but who were treated by DM-P4P-participating physicians were significantly more likely to receive 3/7 of the recommended tests/examinations lipid profile [adjusted RR = 1.24 95%CI = (1.04–1.45)] ($p < 0.05$), ALT [adjusted RR = 1.06 95% CI = (1.00–1.11)] ($p < 0.1$) and eye examination [adjusted RR = 1.21 95%CI = (1.11, 1.31)] ($p < 0.01$) than those treated by non-P4P physicians.
LeBlanc et al. (38)	Longitudinal study	Canada NHI model	Secondary Tertiary	P4P Bonus (annually CAN$83.83/patient for completing all indicators). Performance based on physician level	To study the influence of a P4P program for GPs on the glycemic control of diabetes patients (diabetes disease management).	Primary care (not further specified)	Diabetes patients for which a GP claimed the incentive had greater odds of receiving at least 2 glycemic tests per year (OR = 1.92, 99% CI = 1.87–1.96, $p < 0.0001$) compared to incentive not claimed. These odds increased by 56% (OR = 1.56 99% CI = 1.49–1.62, $p < 0.0001$) following the P4P implementation. No difference in glycemia values between incentive claimed (7.4% SD = 1.4) and incentive not claimed (7.5% SD = 1.4) groups.
Lee et al. (39)	Retrospective cohort study: interrupted time series	UK Beveridge Model	Secondary Tertiary	P4P QOF (representing 25% of income). Performance based on practice level	To evaluate if the P4P program resulted in a step change the quality of care (blood pressure and cholesterol controls) for coronary heart disease, stroke and hypertension.	Primary care (general practices)	The P4P program was associated with an initial trend change pertaining to reduction in systolic blood pressure for hypertension patients (−0.83, CI = −1.08, −0.58) (significance at 1%) compared to the period before implementation. These improvements appear to stabilize in the following years.

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
Li et al. (40)	Natural experiment	Canada NHI model	Primary Secondary	P4P Bonus for reaching all 5 targets = $11.000 + bonus for scheduling appointments for eligle patients = $11.000 - TOTAL (maximum ($22.000) <10% annual revenue). Performance based on physician level	To identify the impact of a P4P incentive on the provision of five preventive primary care services (immunizations and screenings).	Primary care (primary care physicians)	P4P had a statistically significant effect on the provision of adult immunizations (0.028 SE = 0.007), Pap smears (0.041 SE = 0.005), mammograms (0.018 SE = 0.005), and colorectal cancer (0.085 SE = 0.007) (significant at 1%) leaving only the effect on toddler immunizations non-statistically significant. Representing an increase of 5.1, 7.0, 2.8, and 57% respectively over the base compliance levels.
Liddy et al. (41)	Cross-Sectional study	Canada NHI model	Secondary Tertiary	FFS (mainly FFS) vs. Blended Capitation (mainly capitation) vs. Salary	To compare different primary care models regarding the adherence to ten evidence-based guidelines pertaining to cardiovascular disease management.	Primary care (not further specified)	Diabetes care: significantly higher for salaried professionals than fee-for-service practices [Adjusted OR = 2.4 (95% CI 1.4–4.2), $p = 0.001$]. Smoking cessation drug prescription: Blended capitation practices significantly more likely than salaried professionals [AOR = 2.4 (1.3–4.6), $p = 0.007$]. Weight management measurements: Blended capitation practices were significantly more likely to measure waist circumference than FFS practices [19 vs. 5%, AOR = 3.7 (1.8–7.8), $p = 0.0006$]. No significant difference between models for chronic kidney disease, dyslipidemia, and hypertension management.
Merilind et al. (42)	Interrupted time series	Estonia Bismarck model	Primary	P4P (2–4% of GPs reimbursement). Performance based on physician level	To compare childhood immunization rates of Estonian family doctors joined and not joined the P4P program.	Primary care (family physicians)	There was an improvement in both groups during the observation period, however doctors joined to the quality system met the 90% vaccination criterion more frequently compared to doctors not joined to the quality system. Doctors not joined to the quality system were below the 90% vaccination criterion in all vaccinations listed in the Estonian State Immunization Schedule.
Norman et al. (43)	Qualitative semi-structured interviews	UK Beveridge model	Quaternary	P4P QOF Bonus for quality targets pertaining clinical care, practice organization and patient experience (representing	To examine how GPs experience the British P4P program regarding its consequences for their professional ethos.	Primary care (general practices)	Professionals' opinions on P4P's effect on quaternary prevention: P4P (QOF) has the potential to medicalize pre-disease states and risk factors raising concern about

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
				25% of income). Performance based on practice level			over-medicalization. The general trend is to introduce medication early on. Incentives have the power to change doctors' behavior and adapt their practices.
Pan et al. (44)	Retrospective cohort study	Taiwan NHI model	Secondary Tertiary	P4P Diabetes Program Performance is based on 4 indicators–final achievement grade places physician in ranking. Top 25% receive additional bonus. Performance based on physician level	To explore the differences in physician continuity of care and survival rates between P4P participants and non-participants diabetes patients.	Medical specialized care (hospitals and clinics)	P4P participation was associated with a higher continuity of care score (COCI) ($\beta = 0.227$ (SE = 0.001) ($P < 0.001$) compared to nonparticipants. P4P participants had a lower hazard ratio HR of mortality 0.43 (95% CI = 0.41–0.44, $p < 0.001$).
Pearson et al. (45)	Cross-Sectional study	USA non-federally employed physicians in private offices AND community health centers throughout the US	Primary Secondary Tertiary	Different levels of capitation	To determine whether four different levels of capitated payment were associated with patient education being included more frequently compared to other payments.	Primary care (not further specified)	The likelihood of visits including patient education for different levels of capitation (95%CI): <25% capitation: OR = 1.00(1.00–1.00); 25–50% capitation OR = 0.77 (0.38–1.58); 50–75% capitation OR = 0.81 (0.53–1.25); >75% capitation OR = 3.38 (1.23–9.30).
Pendrith et al. (46)	Quasi experiment	Canada NHI model	Secondary	P4P (FFS vs. FFS + P4P vs. Capitation + P4P) Bonus [$220 (60%)–$2,200 (80%)]. Performance based on physician level	To compare cervical cancer screening rates among three reimbursement models and to estimate the average and marginal costs of screening/patient.	Primary care (not further specified)	The mean adjusted screening rates per reimbursement ($p < 0.0001$): (FFS + P4P) 7.7% (95%CI = 7.6, 7.7) higher compared to FFS and 2.3% (95% CI = 2.3, 2.3) higher compared to (Capitation + FFS) (Capitation + FFS). 6.2% (95% CI = 6.2, 6.3) higher than FFS. GPs practicing in (FFS + P4P) and (capitation + P4P) had significantly higher screening rates compared to FFS alone.

(Continued)

TABLE 1 | Continued

References	Study design	Country/delivery model	Level of prevention	Reimbursement	Aims of study	Care domain	Primary findings
Rajkotia et al. (47)	Retrospective case control	Mozambique Program sponsored by NGO	Primary Secondary Tertiary	P4P Bonus ($0.10–$11.20 per target per patient). Performance based on practice level	To evaluate the effects of P4P program in two provinces (North and South) on the provision of 18 HIV and maternal/child HIV preventive services compared to input-based financing.	Health facilities (not further specified)	P4P was associated with an increase of 251.6% [$\beta = 9.1$ (SE = 1.3, $p < 0.001$)] in HIV-infected pregnant women receiving therapy in the North and an increase of 194.6% [19.4 (SE 3.8, $p < 0.001$] in the South relative to the control group. P4P program was associated with significant improvements of 14 indicators in the North and 9 indicators in the South achieving similar improvements. Indicators were not sensitive to price, but rather to the level of effort associated.
Serumaga et al. (48)	Interrupted time series	UK Beveridge model	Secondary Tertiary	P4P QOF (representing 25% of income). Performance based on practice level	To access the impact of P4P incentive on the delivery of quality of care and outcomes among UK patients with hypertension.	Primary care (general practices)	No changes attributed to P4P pertaining to: Blood pressure monitoring (level change = 0.85, 95% CI = −3.04, 4.74, $p = 0.669$ and trend change = −0.01, 95%CI = −0.24,0.21, $p = 0.615$). Blood pressure control (level change = −1.19, 95%CI = −2.06, 1.09, $p = 0.109$ and trend change = −0.01, 95%CI = −0.06, 0.03, $p = 0.569$). Treatment intensity (level change = 0.67, 95% CI = −1.27, 2.81, $p = 0.412$ and trend change = 0.02, 95%CI = −0.23, 0.19, $p = 0.706$) P4P had no effect on the cumulative incidence of stroke, myocardial infarction, renal failure, heart failure, or all cause mortality in both treatment experienced and newly treated subgroups.
Sicsic and Franc (49)	Quasi-Natural experiment	France Bismarck model	Secondary	P4P Bonus–maximum €245/target (80% screened). Performance based on physician level	To study the impact of a P4P program on breast cancer screening.	Primary care (general practitioners)	The probability of undergoing breast cancer screening 1.38 % (95 % CI = 0.41–2.35), did not significantly differ following the implementation of the P4P program.

OR, odds ratio; CI, confidence interval; RR, relative risk; SE, standard error.

evaluates the effect of (different levels of) capitated payments (45), another studies a mix of per-diem reimbursement with FFS as an alternative to pure FFS (29), and one other study compares episode-based payments to FFS (25). In these seven studies, FFS reimbursement is used as the benchmark against which other payment models are compared with respect to one or more outcome measures that capture preventive behaviors.

From the 27 studies, the majority (n=16) pertains exclusively to the preventive behaviors of primary care professionals/practices (27, 30, 32, 34–36, 38–43, 45, 46, 48, 49), while the remaining 11 studies pertain to either a hospital setting (25, 29), multiple settings (23, 24, 31, 33, 37, 44) or do not specify the setting (26, 28, 47).

A total of 12 studies focus on chronic disease management (23, 24, 31, 32, 34, 36–39, 41, 44, 48). While 11 further studies consider preventive care such as screenings (28, 35, 36, 46, 49), immunizations (26, 42) or both (27, 30, 33, 40). The two studies pertaining to hospital care in general (25, 29) are labeled under secondary and tertiary prevention. Two studies focus on activities that correspond to primary, secondary and tertiary levels of prevention (45, 47). From the 27 included studies only one study explicitly addresses quaternary prevention (43).

Both per-diem (29) and episode-based payment (25) are only considered by one study each. Neither of these studies, nor our own semi-structured interviews yielded conclusive support for the claim that these types of reimbursement impacted prevention. Echevin and Fortin (29) observe that adding a per-diem fee in 14 departments at a hospital in Quebec (Canada) increased the average length of stay but had ultimately no impact on the delivery of preventive care. Besides this, none of our interviewees is reimbursed on a per-diem basis, therefore no original empirical evidence was collected on this reimbursement through the interviews of our study. Concerning episode-based reimbursement, Cheng et al. (25) conclude that the effect of DRG (diagnosis-related group) payment in 486 Taiwanese hospitals had no significant impact on healthcare preventable adverse outcomes after discharge. As for empirical findings, our interviewed GPs have experience with episode-based reimbursement in primary care, specifically for chronic disease management. Although interviewed GPs are positive about these programs, the difference in type of episode-based reimbursement (hospital vs. primary care) makes it challenging to draw reliable conclusions on this type of reimbursement.

The remainder of the section focusses on the relation between levels of prevention and the better-documented reimbursement systems: FFS (including multiple payment models FFS vs. salary vs. capitation or different blends of FFS) and P4P, respectively. We wish to stress that the included studies vary greatly in design, which affects the extent to which the results may be interpreted as causal or correlative. Clearly, we do not claim that e.g., "the results of cross-sectional studies are by definition correlative" or that "(quasi-)experiments always facilitate causal conclusions". The causal nature of the results is not always clear to the reader of these studies, us as reviewers included. To inform our analyses, we make mention of study designs in our synthesis of prior research and our own primary research below.

FFS, Capitation, and Salary on Primary and Secondary Prevention

Most of the included papers in our review describe FFS-based reimbursement, sometimes in combination with capitation and/or salary-based reimbursement. First, we discuss our findings on FFS vs. salary vs. capitation on primary and secondary prevention.

In a cross-sectional study, Jusot et al. (33) report that FFS is associated with a higher delivery of primary and secondary preventive services (specifically, immunization, and screenings) compared to salary and capitation, suggesting that under FFS, professionals have incentives to increase service volume. These results are coherent with what interview respondents report about FFS incentives. One of the interviewed healthcare purchasing specialists believes that preventive activities that are reimbursed through FFS, such as immunizations, will be stimulated under this reimbursement scheme. Two professionals under FFS acknowledge the incentive to increase production of the reimbursed service as this will lead to increased revenue and cited that when preventive activities, such as patient education, are not reimbursable through a fee, this will act as a disincentive for that type of prevention.

On the other hand, in a longitudinal study and a cross-sectional study respectively, Kiran et al. (36) and Dahrouge et al. (27) find no statistically significant differences between these payment models pertaining to screening (27, 36) and immunizations (27). Both studies suggest that the practice's structure (number of enrolled patients per full-time equivalent GP) and organizational factors (such as working with electronic reminders or team-based care) could be stronger determinants for the delivery of preventive services. Accordingly, lack of time was recurrently mentioned during our interviews (by both salaried professionals and professional under FFS) as a reason for not addressing prevention. Two salaried respondents (one GP and one PT) believe that their provider organization plays a crucial role in stimulating prevention at practice-level by making the necessary resources available for professionals to be able to focus their efforts on prevention, such as extending the length of consultations. Salaried respondents claim they would be open to invest more time in prevention, but the perceived pressure from the provider organization (reimbursed under FFS) to generate revenue is hampering prevention, as one PT illustrates: *"I think it depends on your employer and their vision [...] and whether or not they want to stimulate certain things [...] The fact that my schedule is overloaded is because [employers] have certain ideas on how they want to organize things making them less flexible [...] and this will ultimately compromise quality of care"*. Consistent with this statement, two professionals under FFS demonstrate no desire to increase consultation length (as the reimbursed service pertains to a consultation with a predefined length) nor regard this as an important enough obstacle for prevention that needs to be overcome. GPs reimbursed under a mix of capitation and FFS regard the responsibility placed on them for providing primary prevention as unrealistic. The GP does not think it is feasible to extend consultation length and spend (more) time addressing prevention during consultations, suggesting that in

order to stimulate prevention in healthcare, other entities such as the municipal health services should be made responsible for addressing prevention. This way, GPs can focus on curative tasks and not patient education: "*As a GP I would really like to apply my medical knowledge and since obesity is a big social problem, I think I would be seeing people all day long and discussing how we are going to tackle someone's obesity. Well, I don't think I would want to do that, no.*"

On the other hand, an interviewed GP recently changed reimbursement from the mixed capitation and FFS to a population-based capitated payment regarding the first segment of GP care. According to this GP, this shift removed the incentive for (over)production. As the provider no longer profits from providing more consultations, this led this GP to extend the consultations' length: *"Now I know what I earn per quarter; it no longer depends on how often I see my patients. So, I choose to take more time for my patients because I don't have to see thirty patients a day to earn my living [. . .]. What we notice is that we no longer, or less often, have to book double appointments [. . .]. And that we have just enough time to approach [prevention]. At first, I was skeptical about it, because you feel that you are losing money by not being able to claim your consultations. But if I compare my practice's finances with those from practices under the traditional reimbursement, I realize that we are definitely not in a bad position financially."*

FFS, Capitation, and Salary on Tertiary Prevention

Concerning disease management, Kiran et al. (36) found that GPs' reimbursement with a greater percentage of FFS presented the lowest improvements in diabetes management while reimbursement mostly composed out of capitated payments achieved the largest improvements in diabetes care. With reference to our own empirical research, one respondent PT believes that FFS hinders prevention by reimbursing professionals for every service provided but with no further incentive to avoid disease development, explore potential risk factors that might be the underlying reason for the patient's health complaint or prevent deterioration of a health condition. Similar to Kiran et al. (36), Liddy et al. (41) observe in a cross-sectional study that practices under FFS showed the greatest gaps in adherence to evidence-based guidelines pertaining to cardiovascular disease care. Capitation and salary were similar to each other in results; While salaried GPs scored significantly higher on glucose level control, capitation was linked to increased weight management and smoking cessation drug prescription when compared to FFS and salary. Pearson's et al. (45) cross-sectional study established that GPs for whom >75% of reimbursement consisted of capitation relative to FFS were three times more likely to provide patient education. In sum, salary, and even more so capitation, rather than FFS, appear to be related to better disease management.

FFS, Capitation, and Salary on Quaternary Prevention

Our rapid review yielded no results on quaternary prevention under FFS, salary nor capitation. Nevertheless, our interviews suggest that overmedicalization is still prevalent in healthcare. When asked about overmedicalization, respondents under these three reimbursement schemes believe that it is mostly driven by patient demand, not by reimbursement, and claim they run responsible practices as overprovision might have consequences for the patient's health and healthcare expenditure: *"We are always critical about what is necessary, what is medically indicated."* However, all professionals acknowledge that in some circumstances they might (partially) give in to patients' demands, as a salaried GP illustrates: *"I also try to negotiate a little bit [...] but yeah, I'm not saying I don't do it. Because you also have a future with that patient, in your doctor-patient relationship. [...] I never give [prescription for blood test] without explaining very clearly what you can and what you cannot get with it [...] Because some things just have consequences [...] and then you enter into an unnecessary medicalization process."*

P4P on Primary and Secondary Prevention

In the remainder of this section, we present our findings on P4P-based reimbursement in relation to the levels of prevention. First, we discuss primary and secondary prevention.

In an interrupted times series study, Chien et al. (26) examines the effect of a piece-rate P4P bonus for full and timely childhood immunization. Results show that immunization within the P4P program increased at a significantly higher rate than the comparison group. Similarly, Merilind et al. (42) reveals that GPs under P4P achieved the target of 90% coverage rate for all vaccinations while the comparison group only achieved the target rate for one vaccination. Both studies suggest that P4P schemes with a bonus specifically for immunizations can improve immunization rates. Pendrith et al. (46) observed that FFS on its own provided low incentives for the delivery of cancer screening and that a P4P bonus for achieving 60% or 80% screening rates for three types of cancer combined with FFS lead to an increased provision of these screenings. The combination of capitation and the same P4P bonus also presented higher screening rates in comparison to FFS, leading authors to suggest that adding these P4P incentives was associated with higher cancer screening rates. Conversely to these findings, Kiran et al. (35) observed that despite the increase in billing for self-reported provision of cancer screenings leading to larger expenditures there was little or no significant increase in cancer screening rates after the introduction of P4P bonuses for the achievement of different targets in screening rates. Similarly, two other studies observed that cancer screening rates (30) and immunization rates (30, 40) did not suffer significant changes after implementation of a P4P bonus among GPs for the achievement of targets pertaining to the delivery of these preventive services. Both studies hypothesize that these incentives representing 3–4% (30) and <10% of annual income (40) might have been too small to induce the desired changes in practice. Besides this, authors suggest that other aspects such as provider training (30) and lack of provider reminder systems (35) could impact performance. Li et al. (40) question P4P's effect on the quality of (preventive) care and suggest that the introduction of five different indicators simultaneously might decrease the likelihood of physician's response to any of them. Further research on why and how P4P's design features can help increase professionals' response is required (40).

Different studies provide different interpretations of how P4P's components influence professionals' performance. De Walque's et al. (28) results reveal that P4P higher incentives (US$4.59) had a greater and significant impact on indicators such as couples HIV testing, compared to lower incentives (US$0.92) for individual HIV testing. The latter shows little or no significant effect on professionals' performance. In line with De Walque et al. (28), Sicsic and Franc's (49) study observes little impact of a P4P program among French GPs on breast cancer screening rates, concluding that the "low-powered" financial incentive (maximum €245/target) did not have enough leverage to stimulate providers. Contrastingly, Rajkotia et al. (47) propose that practices are not necessarily more responsive to more profitable indicators (such as the survival rate after treatment of HIV-infected children with a $11.20 reward) than to less profitable ones ($4.20), but instead prioritize targets that can be achieved with a lower level of effort, in this case the number of HIV-infected pregnant women initiating antiretroviral therapy ($10 reward) or the number of family planning consultations given to HIV-infected women ($5 reward). Taken together, these studies suggest that both bonus magnitude (28, 30, 35, 40, 49) and required effort (30, 47) are important components in a P4P scheme. Our interviews did not produce results pertaining to the effect of P4P on primary and secondary prevention.

P4P on Tertiary Prevention

Regarding the effect of P4P on disease management, two studies reveal that Taiwanese diabetes mellitus (DM) patients of P4P-enrolled GPs had higher continuity of care and lower mortality rates (44) and were more likely to receive the P4P-rewarded guideline-recommended DM examinations than patients treated by non-P4P GPs, as presented in a cross-sectional study by Lai and Hou (37). Two longitudinal cohort studies on cardiovascular disease management (23) and diabetes management (32) observe that financially incentivizing disease management check-ups and treatments also resulted in fewer (avoidable) hospitalizations. Chen et al. (23) suggest that P4P success might be due to an easily achievable target concerning the percentage of patients receiving improved quality of care and reaching positive health outcomes for cardiovascular disease. The low baseline rate for improvement (42%) stimulated participation in the P4P program. Hsieh et al. (31) observe that rewarding professionals for process indicators (e.g., control visits and cholesterol and glucose testing) led to no difference in the number of visits nor tests performed. When outcome indicators rewarding improvement in clinical levels (e.g., cholesterol levels) were added to the P4P program, quality of care improved.

Serumaga's et al. (48) interrupted time series study found that rewarding blood pressure control and drug prescription for hypertension disease management did not increase the delivery of these services in a clinically or statistically significant manner, nor were there changes in mortality rates or other hypertension related adverse outcomes. The authors suggest that blood pressure control had already improved before the implementation of P4P and that P4P targets might have been set too low for significant change to take place. Similarly, the results of both Lee et al. (39) and Chen et al. (24) show that the delivery of secondary and tertiary preventive services did not significantly vary between the P4P and non-P4P groups. Chen et al. (24) cited the small bonus size ($3-$30/service/per patient) and low provider participation as probable explanations for low behavioral change in the delivery of three guideline-recommended preventive services and test for Hepatitis B and Hepatitis C patients. Lee et al. (39) stated that future P4P programs rewarding blood pressure and cholesterol level controls should include achievable but at the same time challenging targets to create enough leverage among different practices.

Two of our interviewed GPs believe that performance targets set by the healthcare purchaser limit their professional autonomy and control the way care is delivered without taking other factors into consideration: "*The healthcare purchaser imposes targets on me that I must meet, which may not be feasible at all for a great part of my patients because I have many elderly people or immigrants, for example, and then I have a problem.*" Another GP adds: *"If I prescribe expensive medication once, it is probably because there is medical necessity, which is never accepted (by the purchaser), because then there are again twenty-six conditions that it must meet."* The interviewees also believe these targets are set primarily in order to reduce healthcare costs and not to increase quality.

In a P4P program financially rewarding providers for conducting all recommended diabetes management actions, LeBlanc et al. (38) conclude that although patients of GPs claiming the bonus received more glucose tests per year and had consequently better GP follow-up, this was ultimately not translated into lower glucose levels for those patients relative to the comparison group. Similarly, Karunaratne et al. (34) prospective longitudinal cohort study examines the management of chronic kidney disease in primary care before and after implementation of P4P bonuses for initial and ongoing management actions such as blood pressure measurement and control. Contrary to Serumaga's et al. (48) findings, in this case blood pressure measurement and prescription medication significantly increased as did costs associated with increased prescribing. However, Karunaratne et al. (34) cannot establish if these events consequently resulted in improved health outcomes, delayed disease progression or decreased mortality. Similarly, in our empirical research, two respondent GPs question the quality improvement incentive P4P programs aims to induce and raised some concerns regarding such incentives of P4P: *"You can also ask yourself whether that [P4P] really improves quality, because you mainly measure whether people have been seen [by the doctor], but whether you will take action or do something with those [blood test] results is the real question."*

P4P on Quaternary Prevention

Norman's et al. (43) interview study with GPs is the only study in our research that explicitly investigates P4P's impact on quaternary prevention. GPs under P4P revealed experiencing an incentive to reduce any risk factor and prophylactically treat patients as otherwise it might disturb P4P target achievement. Also, Norman et al. (43) report that GPs acknowledged opting to medicate patients rather than trying non-pharmacological approaches simply to be able to timely achieve P4P targets, and additionally acknowledged that even when indicators went

against GPs' inner values, they still complied and strived to achieve targets. These reported incentives are acknowledged by two of our own respondent GPs who disagree with the implementation of P4P and worry that money may become the incentive for action, adding: *"Because then we will do things because you get money for it and not because you actually want to work that way"*.

DISCUSSION

Our research collected empirical evidence on the relationships between different reimbursement schemes and four levels of prevention. Most of our results relate to P4P, FFS, capitation and salary. We also obtained results on both per-diem and episode-based payment, however neither the rapid review nor our own empirical research could confirm the impact of these types of reimbursements on prevention. Quaternary prevention was addressed by one study only. The integration of findings from both the rapid review and our original empirical research allows us to draw the following main discussion points regarding reimbursement schemes and prevention.

Salary

The findings of our rapid review on the impact of salary-based reimbursement are ambiguous, associating salary to both higher and lower delivery of preventive services compared to FFS and capitation (33, 41). In previous literature, while Gosden et al. (50) claims salaried professionals want to minimize their personal efforts, Kane et al. (4) proposes that these professionals could be incentivized to engage in preventive care more than professionals reimbursed under FFS. An obstacle experienced by salaried respondents are the incentives from the employer organization and not making the necessary resources available to stimulate prevention, suggesting that the reimbursement of the provider organization or practice might interfere with salaried professionals' behavior toward prevention. Therefore, it should be taken into consideration that these different interactions between the professional and the provider organization can align or misalign incentives which might impact prevention in practice.

FFS

Concerning FFS, although ambiguous, results suggested that its widely reported incentive to increase production might work in favor of preventive services such as immunizations, eye exams and screening for cancer (33). The corollary is that non-reimbursed activities, longer consultations or less consultations might evoke feelings of loss for these professionals, according to empirical findings. In fact, interviewees acknowledged the fact that when prevention is not as widely reimbursed in fees this poses as an obstacle for the provision of preventive services. More than a decade ago, Ellis and Miller (14) already proposed that activities not reimbursed through fees (such as preventive counseling) can be neglected under FFS. Results also suggest that FFS restricts the delivery of care to predefined standards and does not allow the flexibility to organize care delivery differently, as some forms of prevention might require. Therefore, FFS might work for some forms of prevention, in particular when the prevention activity can be specified as reimbursable under FFS (such as immunizations), but it can be questioned whether it will stimulate professionals to address prevention when the preventive activity requires efforts/services that are not reimbursed through a fee.

Population-Based Payment

Both PTs and GPs state that, as the patient's medical complaint receives primary attention during consultations and sometimes the consultation is even too short to address the complaint in detail, there is usually not enough time left to address risk factors and address secondary and tertiary prevention. However, opinions of GPs under population-based capitated payment contrast with those of GPs under traditional capitation and FFS reimbursement. A GP under traditional reimbursement considers the responsibility to deliver preventive services by GPs to be unrealistic and therefore does not feel increasing consultation length to be necessary. Instead, the responsibility for prevention should be placed elsewhere. A focus on revenue and profit might be a reason to increase the number of consultations per day and no desire to extend consultation length. On the other hand, GPs under population-based capitation report having altered the delivery of care to facilitate the delivery of prevention through extending consultation length. This is due to a bigger focus on prevention as a cost reduction strategy from capitation incentives and a reduced volume incentive with the elimination of FFS.

Although respondents show that they are aware of the importance of quaternary prevention, they acknowledge that overmedicalization still persists in healthcare due to patients' demands. In case of time constraints, professionals might be likely to resort to unnecessary prescriptions or referrals in detriment of providing important information, comprehensively discussing alternative approaches that mitigate overmedicalization, compromising quaternary prevention.

P4P

Both our rapid review and our interviews' ambiguous findings question P4P's promise to improve healthcare quality through better prevention. Studies show that the achievement of targets for performing disease management examinations is not necessarily translated into better patient outcomes or effective secondary and tertiary prevention (34, 38). Similarly, Flodgren et al. (51) already concluded in their systematic review that P4P effectively managed to change professional's practice, however no effect on patient outcomes is subsequently observed. Empirical results also report how P4P's metrics might steer professional's behavior and how this might conflict with what is best for the patient. The risk of losing a bonus might trigger professionals to circumvent factors standing in the way of achieving targets. The results of our rapid review underscore the importance of different P4P design features in stimulating behavior. However, the role of bonus size and level of effort in responsiveness to an indicator are discordantly described. While Gavagan et al. (30), Li et al. (40), De Walque et al. (28), Chen et al. (24), and Sicsic and Franc (49) cite the importance of bonus size, Rajkotia et al.

(47) claim that the level of effort necessary to reach a target is the most important determinant of behavior. Serumaga et al. (48) and Chen et al. (23) claim that targets should be low enough to motivate professionals while Chen et al. (24) and Lee et al. (39) report that, in order to make P4P cost-effective, targets should not be set too low and easy to achieve. On the other hand, Hsieh et al. (31) claim that only when incentives for outcome indicators are added to the P4P program, improvements in quality of care are observed. Taken together, these studies suggest that bonus magnitude, required effort and type of indicator are important components in a P4P scheme. Even though our research did not further investigate the role of these components, our results suggest this should be considered as it most likely will influence a reimbursement's effectiveness toward prevention.

Regarding quaternary prevention, Norman et al. (43) show how P4P can lead professionals to resort to (over)medicalization to achieve targets, compromising this level of prevention. Karunaratne et al. (34) show that P4P leads to a rise in medication prescription and costs related to increased prescription with no further improvement in health outcomes. In line with these results, interviewees are critical about P4P and worry that money may become the incentive for action, and this may "crowd out" intrinsic motivation to deliver efficiency and quality in healthcare. While P4P is promoted by purchasers, it is considered disruptive by professionals, who suggest it might trigger different unintended behaviors in professionals that ultimately hinder effective prevention at different levels.

Per-Diem and Episode-Based Payment

Both per-diem and episode-based payment are only considered by one study each and neither can claim these reimbursement schemes impact prevention. None of our interviewees has experience with per-diem reimbursement. Interviewees stated that primary care episode-based payment creates incentives to better organize disease management and actively monitor patients so as to avoid complications from which the provider could incur additional costs. This type of episode-based payment is a form of prospective reimbursement, with revenues known upfront and hence there is a strong incentive for cost avoidance.

CONCLUSIONS

The benefits of preventive health services are widely recognized. However, delivery of prevention services encounters numerous obstacles in healthcare. It is argued that reimbursement schemes play an important role in both hindering and stimulating the provision of healthcare services (13). Nevertheless, there has been little focus on how reimbursement schemes could specifically contribute to the delivery of preventive health services.

Our research provides insights into how different types of reimbursement (e.g., fee-for-service, or pay-for-performance) impact healthcare professionals' behavior; stimulating or hindering their efforts to address prevention. We distinguish between four levels of prevention, ranging from avoiding disease onset, allowing early diagnose and reducing disease impact to protecting patients from receiving redundant, unnecessary care. We find that not one ideal reimbursement scheme exists, providing incentives that stimulate (or hinder) prevention at all its levels. There are, however, certain types of reimbursements that work well for certain types of preventive care services. For example, the volume incentive from FFS could be beneficial for some levels of prevention when clearly specified preventive actions are concerned (such as immunization, as an example of primary prevention or screenings as an example of secondary prevention). On the other hand, population based capitated reimbursement might facilitate the delivery of some forms of prevention that are more difficult to specify as a reimbursable service, or for which lack of time poses as an obstacle under other reimbursement schemes, allowing the flexibility to alter the delivery of care. We also discuss P4P, as this is prominent in both our literature review as well as amongst our interviewees. As our study empirically reported, P4P's incentives might have unintended consequences for professionals' intrinsic motivation. P4P's criteria for medication prescription is an example of how what is being measured and therefore reimbursed for could influence a professional's practice away from what they initially intended. Additionally, the achievement of P4P's targets does not always imply better health outcomes (34, 38). Besides this, our study also describes how the pressure to (timely) achieve targets arising from this type of reimbursement can lead professionals to resort to (over)medicalization and discard other approaches that could better fit the patient's needs, compromising quaternary prevention (43).

Strengths and Limitations

The strength of our study lies in the incorporation of evidence from both a rapid review of the literature and interviews with professionals to help consolidate results and achieve a more comprehensive explanation of how reimbursement schemes can affect prevention. Furthermore, this research raises awareness on overmedicalization by contemplating quaternary prevention as the fourth level of prevention, contributing with a more overarching definition of prevention. Although efforts were made to mitigate bias, this research has a few limitations which will be outlined in this section. These limitations could provide additional guidance for future research.

First regarding the rapid review. Despite efforts to test and strengthen our search strategies, it is entirely possible that studies to prevention have unintentionally been omitted, due to them not being described with the term "prevention." Quaternary prevention is still a relatively new concept and therefore strategies to reduce medical overuse might not be perceived and labeled as (a level of) prevention. In addition, despite the advantages of rapid review methodology, drawbacks must be acknowledged, i.e., the process of data collection, selection and analysis was primarily performed by one reviewer. This may have compromised the study selection procedure and consequently the reliability of results. Also, unlike more traditional systematic literature reviews, rapid reviews rely on narrative analysis and synthesis rather than meta-analysis of the included studies (18). Although we argue that our rapid review fits our research aim well, a more traditional systematic approach would be better suited for more detailed and quantitatively sophisticated meta-analysis of the effectiveness of payment models in relation to e.g.,

case mix of patients. Meta-analysis would also allow for further assessment of the quality of the results and the level of evidence provided by the included studies.

As addressed by some studies included in our review, the effectiveness of any reimbursement scheme is likely to be affected by the generosity of the payment and not solely the type of reimbursement. However, not all studies from our rapid review disclose the reimbursed amounts in analogous ways, making it impossible to compare payment generosity and draw conclusions on this matter. Therefore, we did not investigate this matter in our review which could have provided additional and valuable insights to our research. We recognize this as a limitation of our study.

We acknowledge that there may be various other factors that interact with reimbursement scheme to impact delivery of prevention services. These can be factors related to the healthcare delivery model of a country, the level of investment in healthcare in a country, social, economic or cultural differences between countries, or differences between countries or regions in support structures offered to healthcare professionals other than reimbursement. We summarize the evidence at a relatively high level of abstraction, between reimbursement scheme and delivery of prevention services only, and cannot account for all differences between countries. Moreover, our rapid review is skewed toward studies executed in OECD countries, more specifically North America, and countries with a National Health Insurance (NHI) model. Jusot et al. (33) have shown, in their 14-country study, that factors related to reimbursement were most strongly related to utilization of preventive services, with other system-level factors, like capacity or structure, playing a lesser role. This lends credibility to our assumption that reimbursement scheme is a very important factor, even if it may interact with other factors. Still, looking at reimbursement separately is a limitation, and we recommend future research to also study interactions with other factors.

Concerning the empirical research, interviews were planned to be conducted face-to-face, however, due to local restrictions regarding the COVID-19 outbreak at the time of this study's data collection, interviews had to be conducted via telephone. Furthermore, some of the initially targeted respondents had to cancel their participation. The initial intention was to conceptualize a "theoretical sample" by means of literature review and subsequently select respondents based on that theoretical sample. Both research setting and respondents had to be rearranged in a short period of time and respondents had to be recruited through "convenience sampling" and "snowball sampling" (52), limiting the opportunities to draw a varied sample of respondents as initially intended. Medical specialists and patients, who could have added valuable insights, were not interviewed. Due to the qualitative research design and convenience sampling of a limited number of respondents, this research could lack representativeness and external validity.

Neither the rapid review nor the empirical research provided ample insights on all relevant reimbursement schemes. On the one hand because our sample did not include respondents who experienced all types of reimbursement, and secondly because the rapid review identified relatively many studies on certain reimbursement schemes but less so on others. Future research should target also other respondent samples for a more comprehensive understanding of how reimbursement may affect prevention. Besides this, most respondents were paid under a mix of reimbursements which makes it difficult to assess their isolated effect and can compromise results.

The fact that healthcare professionals' behaviors might be stimulated or hindered by incentives from different types of reimbursement schemes could be regarded as in conflict with the oath of ethics concerning non-maleficence. During the interviews it was noticeable that some respondents were more hesitant to talk about possible incentives altering their behavior in terms of under- and/or overprovision of care and might have held back valuable information.

Finally, our rapid review identified only one study on quaternary prevention and more research is needed on this level of prevention and how different reimbursement schemes impact quaternary prevention.

AUTHOR CONTRIBUTIONS

EZ conceived the idea, performed the review and interviews, and wrote the first draft of the manuscript. ER and HE assisted the rapid review and contributed to the data interpretation process. All authors wrote and edited the manuscript and approved the submitted version.

ACKNOWLEDGMENTS

We thank all the participants in this study. An earlier version of this paper has been presented at the HSMO ScienceClub at Erasmus University Rotterdam. We thank the participants for their constructive comments. We would especially like to thank Dr. Jeroen van Wijngaarden, Pieter Vandekerckhove, and Dr. Sandra Sülz for their constructive and insightful feedback on the manuscript leading to the improvement of our work.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.750122/full#supplementary-material

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APPENDIX

Topic List for Interviews

Introduction

- Thank respondent for their availability
- Briefly describe the purpose and logistics of this interview
- Ask for permission to record interview for the purpose of transcription
- Start the interview by first collecting the respondent's professional information.

Views on prevention

- Professional's definition of prevention
- Prevention in (daily) practice
- Roles in the provision of prevention
- Prevention and the patient

Reimbursement and prevention

- Reimbursement of preventive services
- Preventive care programs
- Encouragements/obstacles for the delivery of preventive services
- Healthcare purchaser's role

Quaternary prevention/Medicalization

- Benefits/risks of prevention for the patient
- Overmedicalization in healthcare
- Causes for overmedicalization
- Healthcare professionals and overmedicalization
- Strategies to mitigate overmedicalization

Conclusion

- Guarantee respondent's anonymity
- Ask permission to use quotes
- Thank the respondent.

frontiers in Public Health

DATA REPORT
published: 28 October 2021
doi: 10.3389/fpubh.2021.708402

The Use Patterns of Medicaid Home and Community Based Services Among Medicare/Medicaid Beneficiaries With Dementia

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Keywords: Medicaid home and community based services, Alzheimer's disease and related dementias, Medicare-Medicaid dual eligible beneficiary, Medicaid policy, race, neighborhood socioeconomic status

OPEN ACCESS

Edited by:
Steven W. Howard,
Saint Louis University, United States

Reviewed by:
Li-Nien Chien,
Taipei Medical University, Taiwan
Louise Meret-Hanke,
The Pennsylvania State University (PSU), United States

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Specialty section:
This article was submitted to Aging and Public Health, a section of the journal Frontiers in Public Health

Received: *17 May 2021*
Accepted: *29 September 2021*
Published: *28 October 2021*

Citation:
Yan D, Wang S, Temkin-Greener H and Cai S (2021) The Use Patterns of Medicaid Home and Community Based Services Among Medicare/Medicaid Beneficiaries With Dementia. Front. Public Health 9:708402. doi: 10.3389/fpubh.2021.708402

INTRODUCTION

Older adults with Alzheimer's disease and related dementias (ADRD) often have high care needs (1). As their care needs increase, many are placed in nursing homes (NHs), even if they would prefer to live in the community (2). The Medicaid home and community-based services (HCBS) program provides health and supportive services that may allow Medicaid beneficiaries with cognitive and physical impairments to maintain living in the community, delaying or preventing NH placement (3). The HCBS cover services such as durable medical equipment, transportation, hospice care, residential care, personal care, home health, and other waiver services (4). As older adults with ADRD have different care needs than other populations, their utilization patterns of these services can be unique. However, to date, the extent to which these services are differentially used to help maintain community living among older adults with ADRD is not known. Furthermore, while prior studies have noted the existence of racial differences in care needs and individual preferences among persons with ADRD (5, 6), it is unknown whether differences in HCBS utilization patterns between Black and white service users with ADRD also exist. Lastly, it is unknown whether the patterns of HCBS utilization vary by the socio-economic status of the community, which can be closely related to racial differences in individual health, care needs, and ability to maintain community living.

Therefore, the main objective of this study is to address these gaps in knowledge. More specifically, we explored the differences in the pattern of Medicaid HCBS utilization among Black and white Medicare-Medicaid dual eligible older adults with ADRD, and how such racial differences varied by the socio-economic status of the community in which an individual resides. This is an important question to address in order to identify services that may be potentially under-utilized, so as to better target services to the needs of the population.

MATERIALS AND METHODS

Data

The Medicaid Analytic eXtract (MAX) Personal Summary (PS) and Other Therapy (OT) files were obtained for all eligible individuals in the U.S. between 2010 and 2012, and individuals in 28 states in 2013 (due to the availability of data at the time of the data request). These data were then linked with the following 2010–2013 dataset at the individual level: Minimum Data Set (MDS) 2.0/3.0, Medicare Master Beneficiary Summary File (MBSF), and Medicare Provider Analysis and Review (MedPAR). The MAX PS and OT files include the utilization and expenditure

of all types of HCBS for each Medicaid enrollee. The MDS contains information on individuals' NH placements for those admitted to Medicaid- and/or Medicare-certified NHs. MBSF contains information on individuals' demographics, chronic conditions, and death date information for Medicare beneficiaries. The MedPAR file contains information on hospitalization events for Medicare beneficiaries.

Study Population

We initially included 1,758,640 new HCBS users with ADRD who were dually eligible for Medicare and Medicaid and started to use HCBS between February the 1st 2010 through January the 1st 2013. Medicare and Medicaid dually eligible fee-for-service (FFS) beneficiaries were identified using MBSF and MAX PS files. We excluded the Medicare/Medicaid managed care enrollees because their HCBS utilization and hospitalization data were not available (8.6% of the sample was excluded). Diagnosis of ADRD was based on the MBSF chronic condition files. The diagnosis of ADRD in the MBSF chronic condition file was determined based on the ICD-9 codes of all Medicare claims within the past 3 years (7). New HCBS users were defined as those who did not have HCBS episodes in the prior 30 days, based on the OT records. If an individual had multiple eligible episodes over the study period, we only selected the first HCBS use. In total, the final analytical sample included 1,164,225 individuals.

Variables

The outcome variables included the utilization of each of the following HCBS service categories within 365 days of the first state date of HCBS (i.e., the follow-up period): durable medical equipment, transportation, hospice care, residential care, personal care, home health, targeted case management, adult day care, private duty nursing, and other waiver services. Each service type was defined as dichotomous, indicating whether such services had been used or not during the follow-up period. These service categories were identified based on MAX OT file Community Based Long-term Care (CLTC) flag and included both the state plan services and the waiver services (4). These services account for more than 95% of Medicaid HCBS spending.

Key variables of interest were race and neighborhood socioeconomic status. The race of beneficiaries was dichotomized as white or Black using the MBSF Research Triangle Institute (RTI) race variable (8). Neighborhood socioeconomic status was determined based on the 2015 Area Deprivation Index (ADI) (9). The ADI is a validated, neighborhood-level composite index reflecting 17 social determinants of health such as income, education, employment, and housing quality. Its rankings range from 1 to 100, with more disadvantaged neighborhood conditions designated by a higher score. We dichotomously defined disadvantaged neighborhoods as those with an ADI score >55, which was the average ADI score of the study sample.

We also included several covariates, including age, gender, years since first diagnosis of ADRD, chronic conditions (e.g., diabetes, depression, cardiovascular disease, cancer, etc.), and county-level HCBS intensity defined as Medicaid spending on HCBS per user per month.

Analysis

All analyses were conducted at the individual level. We firstly fit a set of linear probability models with county fixed-effect and robust standard errors to examine the relationship between race and the use of each type of HCBS without accounting for other covariates. The linear probability model approximates the logit model and provides the direct interpretation of the coefficients (i.e., change in the probability of outcomes given one unit change in an independent variable) (10, 11). In these models, the coefficients of the race captured overall racial differences in the probability of using each type of HCBS between Black patients and white patients. We then estimated a set of models by adding additional individual characteristics (e.g., age, gender, diagnosis years of ADRD, chronic conditions) and county-level HCBS intensity to explore how much of the overall racial differences could be explained by these variables. Lastly, we stratified the analyses by the socio-economic status of the neighborhood (i.e., whether a community was economically disadvantaged or not, based on ADI score) and examined whether the racial difference in the pattern of HCBS utilization varied with these two types of neighborhoods (12). Lastly, although not included in the main analyses, we also compared the pattern of other health care utilization, including hospitalization (based on MedPAR data) and nursing home placement (based on the MDS data), and any mortality within 365 days of HCBS initiation date.

All analyses were performed using SAS 9.4 (SAS Institute Inc.) and STATA 16 (StataCorp LLC. College Station, TX). This study has been reviewed and approved by the Research Subjects Review Board. The final dataset was saved in SMDNAS College-based data storage. All authors have no conflicts of interest.

RESULTS

Among the analytical sample, 79% of care recipients were white and 21% were Black. The annual spending on HCBS services was higher among white beneficiaries than among black beneficiaries ($5,939 vs. $5,163, $P < 0.01$). **Table 1** compares the pattern of HCBS use and individual characteristics by race. Overall, hospice care had the highest median spending ($27,622 per user per year), followed by residential care ($14,695 per user per year) and personal care ($5,241 per user per year). Personal care had the longest median duration days among HCBS users (152 days per user per year) followed by other waiver services (82 days per user per year) and home health (25 days per user per year). In addition, Black recipients were generally younger, but the distribution of chronic conditions was mixed–for example, Black patients were more likely to have chronic kidney disease, stroke, and diabetes, but were less likely to have depression, anxiety disorders, and osteoporosis than their white counterparts. White HCBS users were also more likely to have NH placements than were Black users. Among those users without any NH placement within 1 year of HCBS use, Black users had a higher hospitalization rate but a lower mortality rate than white users (more details shown in **Table 2**).

Figure 1A shows the unadjusted probabilities (i.e., without controlling for other covariates) of using each type of HCBS

TABLE 1 | Distribution of HCBS use and individual characteristics by race.

	Race		
	White	**Black**	**All**
Number of individuals (%)	916,422 (78.72%)	247,803 (21.28%)	1,164,225 (100.00%)
The penetration of each HCBS type utilization			
Medical equipment	58.58	65.37	60.03
Transportation	40.07	43.39	40.78
Hospice care	14.42	7.61	12.97
Other waiver service	10.83	13.90	11.49
Residential care	6.15	2.16	5.30
Personal care	7.70	10.74	8.35
Home health	5.92	6.29	6.00
Median annual spending on each HCBS type			
Medical equipment	$128	$134	$129
Transportation	$190	$187	$190
Hospice care	$27,827	$26,330	$27,622
Other waiver service	$2,570	$2,875	$2,632
Residential care	$14,747	$13,792	$14,695
Personal care	$5,104	$6,338	$5,241
Home health	$1,588	$2,176	$1,745
Median duration days of each HCBS type within 1 year			
Medical equipment	2	3	2
Transportation	2	3	2
Hospice care	11	10	11
Other waiver service	83	79	82
Residential care	13	13	13
Personal care	152	156	152
Home health	25	33	25
Number of individuals	916,422	247,803	1,164,225
Individual factors			
Age	77.98	75.10	77.36
(SD)	(13.06)	(13.27)	(13.16)
Years since ADRD diagnosis	2.07	1.66	2.03
(SD)	(1.90)	(1.70)	(1.67)
Average spending on HCBS within 1 year	5938.67	5163.35	5772.75
(SD)	(15452.27)	(12281.15)	(14834.19)
Average monthly spending per HCBS user at county level			
<$700	30.82	30.86	30.83
≥$700 and <1,000	37.44	35.94	37.12
≥$1,000	31.74	33.20	32.05
Female	30.59	34.10	31.34
Living in disadvantaged neighborhoods	48.50	32.70	45.15
Acute myocardial infarction	8.13	6.31	7.74
Chronic kidney disease	36.27	44.27	37.98
Chronic obstructive pulmonary disease	45.22	36.45	43.34
Heart failure	52.93	53.88	53.14
Diabetes	48.12	59.57	50.57
Ischemic heart disease	64.14	62.46	63.78
Depression	64.79	48.18	61.23
Osteoporosis	32.17	16.14	28.74
Rheumatoid arthritis/osteoarthritis	63.89	59.66	62.98
Stroke/transient ischemic attack	33.38	37.84	34.33
Asthma	17.96	18.11	17.99
Cancer	14.07	13.64	13.98
Anxiety disorders	37.56	22.29	34.29
Bipolar	13.16	9.53	12.38
Obesity	16.87	19.72	17.48
Death rate within 1 year	26.69	19.69	25.20

The differences among the two race groups of all variables were statistically significant at 0.01 level.

TABLE 2 | Nursing home placement, hospitalization, and mortality rate within 1 year among HCBS users by race.

	Race			
	White		**Black**	
	N	**%**	***N***	**%**
Any nursing home entry	604,162	65.9%	133,825	54.0%
Community stayer without nursing home entry	312,260	34.1%	113,978	46.0%
Community stayer with hospitalization	87,576	28.0%	36,523	32.0%
Community stayer mortality rate	41,477	13.3%	11,883	10.4%
All HCBS user	916,422	100%	247,803	100%

The differences in the nursing home placement, hospitalization, and mortality rate between blacks and whites were statistically significant at 0.01 level.

among Black beneficiaries vs. white ones. Being Black was associated with a 6.8 percentage point lower probability of using hospice care compared to white individuals ($P < 0.01$), and a 2.1 percentage point lower probability of using residential care, compared with whites ($P < 0.01$). In contrast, Black care recipients had a higher probability of using the medical equipment (5.5 percentage point, $P < 0.01$), personal care (3.0 percentage point, $P < 0.01$), other waiver services (3.2 percentage point, $P < 0.01$), transportation (2.6 percentage point, $P < 0.01$), and home health provisions (0.9 percentage point, $P < 0.01$) than white service recipients with ADRD.

Figure 1B presents the adjusted probability of using each type of HCBS among Black patients vs. white patients after controlling for individual-level covariates and county-level HCBS intensity. The findings were similar to those in unadjusted models: Black patients had a lower probability of using hospice care and residential care, but a higher probability of using the other five types of HCBS than white patients did. The racial differences in using medical equipment (5.5 vs. 7.1 percentage points, $P < 0.05$) and transportation (1.8 vs. 2.6 percentage points, $P < 0.05$) were smaller in adjusted models than those in unadjusted models.

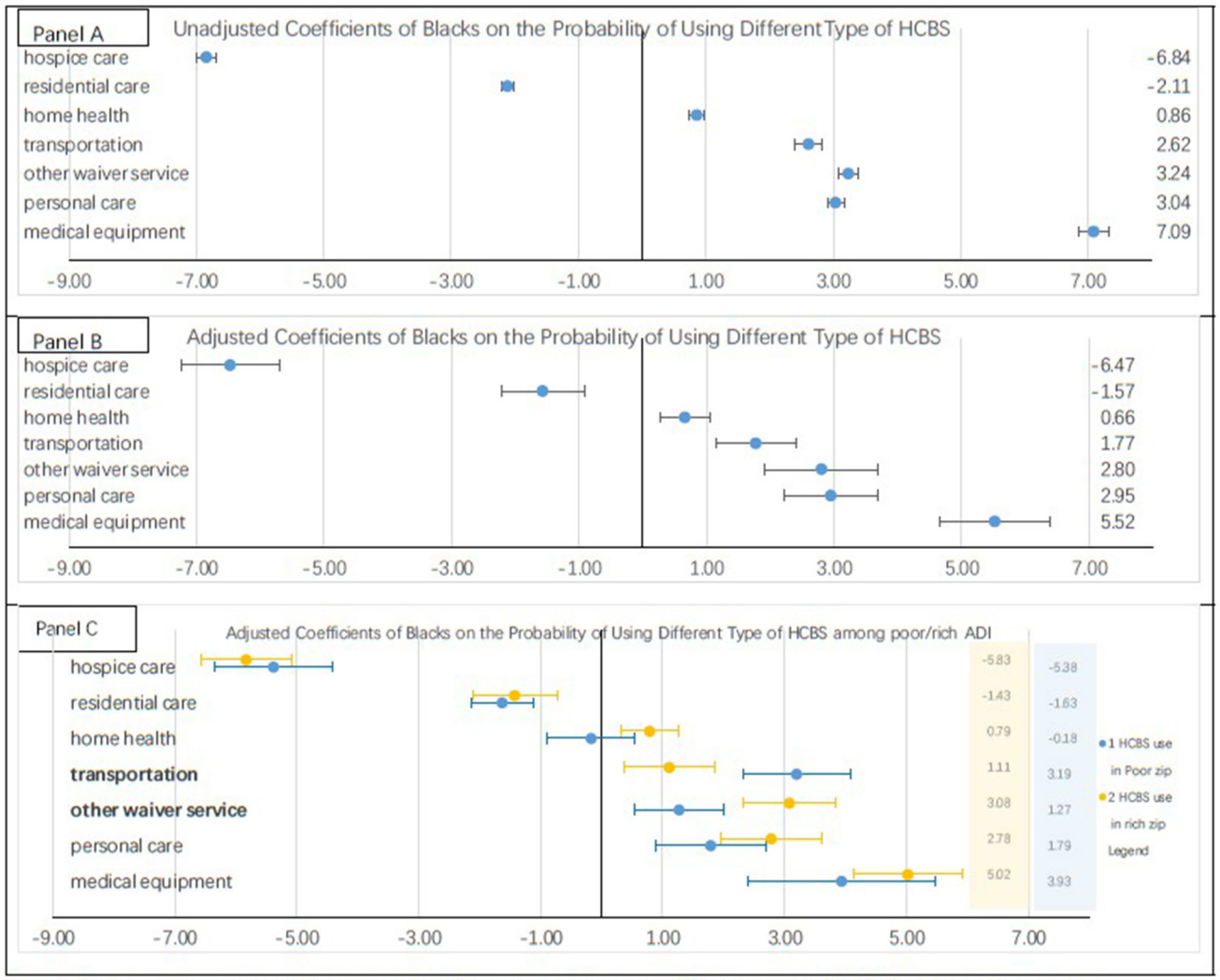

FIGURE 1 | Users' probability of using each type of HCBS. **(A)** shows the results of linear probability regressions on different types of HCBS. The values are the coefficients of blacks on the probability of using different types of HCBS. The regression models added county fixed effect but did not control other variables. **(B)** shows the results of linear probability regressions on different types of HCBS. The values are the coefficients of blacks on the probability of using different types of HCBS. The regressions adjusted individual characteristics (i.e., age, gender, and diagnosis years of ADRD), chronic conditions (e.g., diabetes, depression, cardiovascular disease, cancer etc.), county level Medicaid spending on HCBS per HCBS user per month. **(C)** shows the results of linear probability regressions on different types of HCBS among individuals in disadvantaged neighborhoods and non-disadvantaged neighborhoods, respectively. These regressions adjusted covariates included in the **(B)**. Rich communities are those with 2015 ADI <55. We use 55 as a cutoff point because the mean ADI of the study population is 55.3. With this cutoff 55% of sample was in rich communities.

Figure 1C illustrates racial differences in the adjusted probabilities of using each type of HCBS among those residing in disadvantaged vs. non-disadvantaged neighborhoods, respectively. In both of the stratified samples, Black residents had a lower probability of using hospice care and residential care than did white residents. In addition, Black service users had a higher probability of using medical equipment, personal care, other waiver services, and transportation than did white users. The difference in using transportation between Black and white patients was larger in disadvantaged neighborhoods than that in non-disadvantaged neighborhoods (3.2 vs. 1.1 percentage points, $P < 0.05$). In contrast, the racial difference in using other waiver services was smaller in disadvantaged neighborhoods than that in non-disadvantaged neighborhoods (1.3 vs. 3.1 percentage point, $P < 0.05$).

DISCUSSION

In this study, we examined racial differences in the patterns of HCBS use among Medicare-Medicaid duals with ADRD. We found that the total spending on HCBS services was lower among Black enrolees than that among white enrolees. Black patients with ADRD appeared to use different types of services than their white counterparts. Such differences could not be fully explained by the selected sets of individual characteristics.

Moreover, HCBS use patterns varied by the socio-economic status of the community in which an individual resides.

While we found that Black patients tended to use certain services more than white patients did, it is unclear what may have driven such racial differences. One possible explanation may be related to different care needs between Black and white HCBS users with ADRD. Black HCBS users were generally younger, had fewer years with an ADRD diagnosis, and were less likely to approach the end of life than white users. Thus, they may be more likely to rely on less expensive HCBS (such as transportation and medical equipment) to support their community living than white users. Moreover, the different distribution of comorbidities between white and Black people with ADRD may also contribute to the different patterns of HCBS utilization. For example, Black ADRD patients were more likely to have chronic kidney disease and stroke than white ADRD patients. Individuals with these comorbidities always need transportation to and from the hemodialysis center or often depend on emergency care (13, 14). Therefore, individuals with these comorbidities may be more likely to use Medicaid transportation services. Indeed, after controlling for individual health conditions, the racial differences in using transportation services were reduced.

The findings of this study further suggest that neighborhood socioeconomic status may also influence the pattern of HCBS utilization. For example, Black individuals residing in disadvantaged neighborhoods had a higher likelihood to use Medicaid transportation assistance than Black individuals in non-disadvantaged neighborhoods. This might be related to the availability of relevant medical services in these communities. Studies have suggested that individuals in more disadvantaged neighborhoods may have to drive further to receive specialist care (15) thus leading to higher utilization of transportation services.

Lastly, the detected racial differences in HCBS utilization may also be related to individual preferences with regard to different services. For example, our findings indicate that Black patients are less likely to enroll in hospice than white patients, and such racial differences do not seem to be affected by the set of observed individual characteristics. It is likely that Black service users prefer more intensive treatment due to the historical undertreatment of black patients (16). The Medicaid hospice role is very small among Medicare and Medicaid dually eligible beneficiaries (Medicare and Medicaid accounted for about 74 and 7% of total hospice revenues, respectively) since the Medicaid programs just pay Medicare hospice copayments or some optional service that are not covered by Medicare (17). The total hospice care use may be different from that found in this study.

Several limitations should be mentioned. First, although the study population in this study are Medicare-Medicaid dually eligible, we were not able to examine whether they had barriers, other than insurance status, in access to different types of HCBS services. Secondly, some potential drivers of HCBS use, such as for example personal preferences or the availability of family caregiver support, cannot be ascertained from claims data. Thirdly, this study just included fee-for-service beneficiaries but excluded beneficiaries covered by Managed Care. Therefore, the results of this study may not be applied to those Managed Care beneficiaries. Future research should further examine other potential reasons underlying the difference in HBCS use. In addition, this study did not include Medicare-covered services. However, Medicare and Medicaid have different roles in paying for health services. For example, Medicare home health services are used for post-acute rehabilitative care needs, while Medicaid reimburses for other in-home personal attendant services that are specifically excluded from Medicare coverage (18, 19). Home health services paid for by Medicare and those paid for by Medicaid are different and are not substitutes for each other. Thus, we do not think that focusing on Medicaid HCBS services alone is a limitation. Lastly, our identification of ADRD is based on the Medicare data. Although it is possible that we under-identify the population with ADRD, this is the best data source available to us to identify ADRD population. Despite these limitations, to the best of our knowledge, this is the first study that used national data to examine racial differences in HCBS use patterns and health outcomes among HCBS users with ADRD.

The findings of this study shed light on how HCBS services are used by white and Black duals with ADRD. These HCBS use patterns differences among white and black individuals with ADRD could be important as policymakers target service availability to this population to improve care and delay or prevent institutional care.

DATA AVAILABILITY STATEMENT

According to the CMS requirement of MAX data, we cannot open the raw data of this study to the public. Instead, the raw data will be saved in the College-based data storage according to CMS and University of Rochester Data use policy.

AUTHOR CONTRIBUTIONS

DY, SW, HT-G, and SC contributed to conception and design of the study. DY and SW organized the database. DY performed the statistical analysis and wrote the first draft of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

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frontiers in Public Health

ORIGINAL RESEARCH
published: 17 November 2021
doi: 10.3389/fpubh.2021.707907

A Quasi-Experimental Study of Medicaid Expansion and Urban Mortality in the American Northeast

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Objectives: To investigate the association of state-level Medicaid expansion and non-elderly mortality rates from 1999 to 2018 in Northeastern urban settings.

Methods: This quasi-experimental study utilized a synthetic control method to assess the association of Medicaid expansion on non-elderly urban mortality rates [1999–2018]. Counties encompassing the largest cities in the Northeastern Megalopolis (Washington D.C., Baltimore, Philadelphia, New York City, and Boston) were selected as treatment units (n = 5 cities, 3,543,302 individuals in 2018). Cities in states without Medicaid expansion were utilized as control units (n = 17 cities, 12,713,768 individuals in 2018).

Results: Across all cities, there was a significant reduction in the neoplasm (Population-Adjusted Average Treatment Effect = −1.37 [95% CI −2.73, −0.42]) and all-cause (Population-Adjusted Average Treatment Effect = −2.57 [95%CI −8.46, −0.58]) mortality rate. Washington D.C. encountered the largest reductions in mortality (Average Treatment Effect on All-Cause Medical Mortality = −5.40 monthly deaths per 100,000 individuals [95% CI −12.50, −3.34], −18.84% [95% CI −43.64%, −11.67%] reduction, p = <0.001; Average Treatment Effect on Neoplasm Mortality = −1.95 monthly deaths per 100,000 individuals [95% CI −3.04, −0.98], −21.88% [95% CI −34.10%, −10.99%] reduction, p = 0.002). Reductions in all-cause medical mortality and neoplasm mortality rates were similarly observed in other cities.

Conclusion: Significant reductions in urban mortality rates were associated with Medicaid expansion. Our study suggests that Medicaid expansion saved lives in the observed urban settings.

Keywords: Medicaid expansion, Medicaid, cities, mortality, urban

OPEN ACCESS

Edited by:
Steven W. Howard,
Saint Louis University, United States

Reviewed by:
Rebecca Thorsness,
Brown University, United States
Alyssa Coleman,
Saint Louis University, United States

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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *05 October 2021*
Accepted: *08 October 2021*
Published: *17 November 2021*

Citation:
Ayubcha C, Pouladvand P and Ayubcha S (2021) A Quasi-Experimental Study of Medicaid Expansion and Urban Mortality in the American Northeast. Front. Public Health 9:707907. doi: 10.3389/fpubh.2021.707907

INTRODUCTION

The Affordable Care Act (ACA) offered states the opportunity to expand health insurance coverage to non-elderly adult populations through Medicaid expansion (ME). States were able to use federal funding to increase state Medicaid coverage to all those US Citizens and permanent residents with incomes at or below 138% federal poverty level (FPL) (1). Specific narrow categories of eligibility (e.g., impoverished pregnant women) were federally mandated earlier. Prior to the 2014 implementation of ACA Medicaid expansion, some states utilized waivers to preemptively expand their programs sometimes with more expansive eligibility criteria, but significant gaps in coverage

persisted (2). As a result of Medicaid expansion, Medicaid take-up increased in less-educated, low-income, minority, and younger adults residing in expansion states as compared to peers in nonexpanded states (3). What remains unclear is whether this increase in coverage improved health outcomes, particularly whether urban settings observed reductions in mortality.

Cost-benefit considerations, entailing monetary cost, value of increasing coverage, and the quality of care provided to beneficiaries, have been of notable interest in the debate to increase public medical coverage (4). The original Medicaid program has been associated with moderate decreases in mortality depending on the methods employed (5). Expansions of Medicaid to pregnant women and children during the 1980s were linked to decreases in infant mortality and maternal mortality in most studies (6, 7). Analyses of the 2006 Massachusetts Health Care Reform (MHCR) found reduced all-cause mortality by nearly 8.2 deaths per 100,000 adults (8–10). However, the Oregon Healthcare Experiment study suggested that the effects of coverage may not be immediate or large (11). Such variability may suggest the mechanisms and impact of health coverage is contingent upon alternative factors (e.g., urban-rural residence, minority identity, socioeconomic status, etc.); specific benefits to certain sub-populations or temporally removed effects may further underlie such variability. Smaller mortality effects may also be challenging to capture as gains in public insurance can often be centered in younger populations though older populations are most likely to benefit from coverage.

A number of studies have examined the ACA Medicaid expansion directly and these studies also gesture toward effect heterogeneity in increasing coverage. Nationwide studies observed a decline in all-cause mortality following the ACA Medicaid expansions, but not in cause-specific mortality rates (cardiovascular, respiratory, suicide, and opioid overdose) (12) while a separate analysis found only small, insignificant effects (13). Emerging evidence suggests that Medicaid expansion may have reduced excessive mortality for minorities, in part, by reducing amenable mortality (10, 14). These reforms also seem to have reduced maternal mortality rates, particularly for late-maternal deaths and Black mothers (15, 16). Some studies have observed improvements in specific mortality measures, such as cardiovascular mortality in near-elderly populations and one-year mortality rates among end-stage renal disease patients (17, 18).

We examine the influence of Medicaid expansion on the mortality rates in various Northeastern urban centers. Urban populations warrant targeted study as they are largely distinct from state-wide populations with respect to diversity, healthcare access barriers, types of disease burden and disparities; this is not to mention that a majority of Americans reside in urban settings (19–24). Notably, most large American metropolitan centers are unique in characteristics and state policy history which presents difficulties in studying the generalized "urban" populations. This motivates our study to take a narrow and detailed examination into each city to determine the impact of Medicaid expansion. Accordingly, the observed changes across the included cities may be a means to logically deducing certain city-specific environmental factors that underlie treatment effects. Our study does not only examine all-cause medical mortality but also certain cause-specific mortality rates (e.g., circulatory mortality, neoplasm mortality, etc.). This study will help to understand whether expanding medical coverage can reduce all-cause and cause-specific urban mortality rates, thus helping illuminate why health coverage may improve health for some but not all.

MATERIALS AND METHODS

Study Design and Inclusion Criteria

We utilized a quasi-experimental design to assess the mortality rate among those aged 20–64 from 1999 to 2018; where observed mortality rates after Medicaid expansion were compared to respective predicted mortality rates for each treated city. Only cities in the Northeast megalopolis were considered when selecting treated urban counties. The first year of treatment for each treated city unit was considered the year in which statewide Medicaid expansion was enacted. Urban counties within non-expansion states were selected for the control group. All urban counties with sufficient population levels (>9,000 individuals) and population density and (>800 individuals/mi2) were included. States and counties with any previous Federal Poverty Level (FPL)-based waiver expansions were eliminated from this control pool (e.g., Wisconsin, St. Louis City, MO). The counties utilized in this study can be found in **Table 1**.

Data Sources

All data were secondary, public, and de-identified; no institutional review board approval or informed consent was required. County-level age-adjusted mortality data for individuals between the ages of 20–64 were compiled from the Centers for Disease Control WONDER Tool. The following categories of mortality were included: diseases of the circulatory system mortality (circulatory mortality), diseases of the respiratory system mortality (respiratory mortality), all-cause medical mortality, and neoplasm mortality (i.e., cancer, malignancy). All-cause medical mortality was defined as all-cause mortality absent external-cause mortality. Categories of mortality were defined by International Classification of Diseases (ICD) coding systems; the ICD 9 to ICD 10 code transition was reconciled.

Several longitudinal county-level covariates were obtained and utilized as the basis for developing the synthetic control for each treatment city. From 1999 to 2018, healthcare coverage rates were attained from the Small Area Health Insurance Estimates, non-Hispanic white percentages of populations between 20 and 64 were calculated using the Bridged-Race Population Estimates, inflation-adjusted median income and poverty rates were captured in data from the Bureau of Economic Analysis and the Small Area Income and Poverty Estimates. The Economic Research Services of the United States Department of Agriculture provided educational attainment fractions as defined as the fraction of those with at least some college.

TABLE 1 | Treatment conditions of included cities.

City	County	State	Expansion year	Expansion FPL	Treatment
Baltimore[†]	Baltimore City	MD	2014	138%	Treatment
San Antonio	Bexar	TX	None	None	Control
Fort Lauderdale	Broward	FL	None	None	Control
Dallas	Dallas	TX	None	None	Control
Nashville	Davidson	TN	None	None	Control
Washington D.C.[††]	District of Columbia		2010	210%	Treatment
Atlanta	Fulton	GA	None	None	Control
Houston	Harris	TX	None	None	Control
Kansas City	Johnson	KS	None	None	Control
Charlotte	Mecklenburg	NC	None	None	Control
Memphis	Shelby	TN	None	None	Control
Miami	Miami-Dade	FL	None	None	Control
New York City[‡]	New York	NY	2014	138%	Treatment
Oklahoma City	Oklahoma	OK	None	None	Control
Orlando	Orange	FL	None	None	Control
Philadelphia	Philadelphia	PA	2015	138%	Treatment
Tampa St. Petersburg Clearwater	Pinellas	FL	None	None	Control
Salt Lake City	Salt Lake	UT	None	None	Control
Boston[‡‡]	Suffolk	MA	2014	138%	Treatment
Fort Worth	Tarrant	TX	None	None	Control
Austin	Travis	TX	None	None	Control
Raleigh	Wake	NC	None	None	Control

[†] 07/01/06 § 1115 Waiver that established the Primary Adult Care program to expand coverage (prescription, primary care, behavioral health) to childless adults at or below 116% of FPL.
[††] 07/01/10 State Plan Amendment extends Medicaid coverage to 133% FPL | 12/01/10 § 1115 Waiver Early ACA expansion extends Medicaid program to 210% of FPL.
[‡] 10/01/01 § 1115 Waiver extends Medicaid Family Health Plus to childless adults at 100% FPL.
[‡‡] 4/6/06 Massachusetts implemented reforms to expand insurance coverage to low- income adults beginning in 2006.

Unavailable covariate data for health insurance coverage (1999–2005) were interpolated using only the trends provided by five-year American Community Survey estimates and Census data.

Main Data Outcomes

Mortality was acquired from 1999 to 2018 and all rates were calculated as age-adjusted deaths per 100,000 population of 20–64-year-olds in the county. Monthly data were used for all-cause medical mortality and neoplasm mortality rates, while yearly data were used for all other forms of mortality; more granular data (i.e., monthly as opposed to yearly) provided greater statistical power but less common forms of mortality lacked sufficient prevalence to justify monthly analyses in light of limited sample sizes and elevated risk of stochasticity.

Statistical Analysis

This study employs synthetic control methods which are advantageous when no single control unit can serve as an ideal comparator for a treated unit. This application creates a synthetic control city unit through a weighted combination of the control cities. The synthetic city is intended to simulate the mortality rate of the treated city (e.g., Philadelphia) in the post-treatment period (e.g., 2015–2018) if Medicaid had not been expanded. The particular weights varied in the specific model as applied to each treated city; selection of weights chiefly aims to create a synthetic city with similar pre-intervention covariate characteristics (e.g., inflation-adjusted median income, lagged mortality rates) to the treated city; this algorithmic process employed by the GSC is akin to creating a control city with similar characteristics to the treated city. The appropriateness of the synthetic city as a comparator to the treated city is determined by the convergence of the pre-intervention outcome trends between the treated city and the synthetic city. Such that, any divergence of trends in the post-intervention can be attributed to the intervention (i.e., Medicaid expansion).

In this study, we utilize a Generalized Synthetic Control (GSC) model (25) for each treated city and specific mortality rate. The GSC model relies upon an interactive fixed effects (IFE) technique within the synthetic control framework. The use of IFE incorporates two-way fixed effects which enabled our model to account for unobserved unit-specific and time-specific confounding variables. IFE modeling of the control units is then used to create out-of-sample predictions for the treated unit which results in a GSC output of the synthetic city's mortality rates. The difference between the synthetic and treatment mortality outcomes is considered the treatment effect on the treated unit. The post-intervention difference between the trends can be

averaged over the post-treatment period; termed the average treatment effected in the treated units (ATT). Parametric bootstrapping inference tests were applied to ascertain uncertainty estimates (25).

One sensitivity test was performed by rerunning the models of the monthly data in yearly format to determine whether the structure of yearly analyses influenced the model. An in-time placebo sensitivity analysis was performed applying a false intervention point in the middle of the pre-intervention period and running the GSC model through the pre-intervention period; specifically, the pre-treatment period was considered 1999–2005 and the post-treatment period was considered 2006–2007 so that the 2008 economic downturn was not included. Given that insurance rate is considered a significant mediating variable, a secondary analysis was also performed utilizing county-level insurance rates for each of the treatment cities. The GSC model was applied to insurance coverage rates as a secondary outcome using all the predictors utilized in the main models except for mortality data. All statistical analyses were conducted in R (version 4.0.0) using the gsynth package.

The GSC overcomes some limitations of alternative methods. The GSC does not rely upon the parallel trend assumption that is required in the difference-in-differences methods (25). The GSC method has further proven to be less sensitive to idiosyncratic volatility with a small number of observations (25). Furthermore, this method does not require sensitivity tests of model specifications as a cross-validation procedure selects the optimal number of factors in the IFE model (25). Nevertheless, the GSC model does assume that a stable combination of control units based on the pre-intervention period characteristics of the treatment and control pool can approximate the outcomes of the treated unit in the post-intervention period absent the intervention. Such advantages may achieve similar or superior performance as compared to other methods; several simulated and applied health policy studies comparing IFE, GSC, DiD, and synthetic control methods have found that GSC models perform best (25, 26). Finally, the GSC structure allows for robust parametric bootstrapping inference whereas such quantitative inference is unavailable for traditional synthetic control methods.

RESULTS

We found evidence of significant reductions in all-cause medical mortality as compared to predicted mortality had expansion not occurred in four cities, including Washington D.C., Baltimore, Philadelphia, and New York, after their 2010, 2014, 2015, and 2014 expansions of Medicaid, respectively (**Figures 1**–**5**). Similar

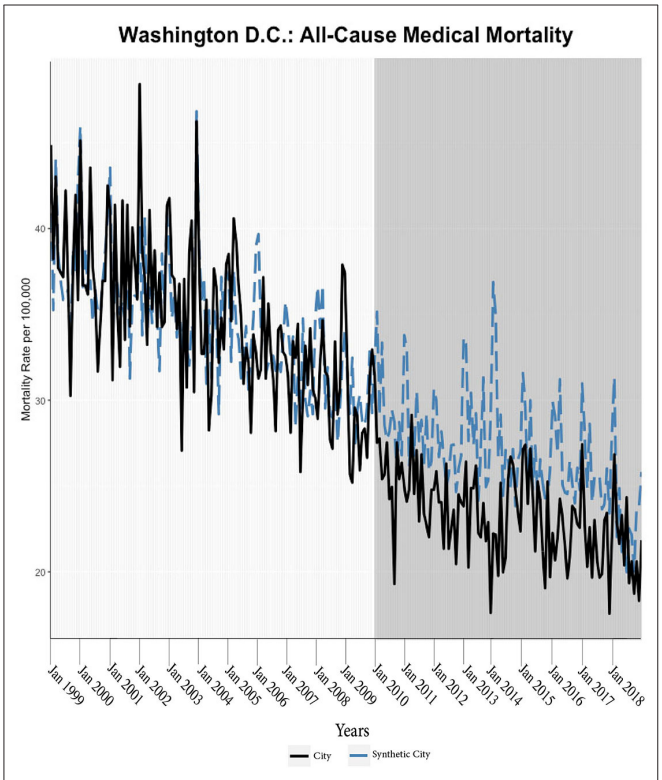

FIGURE 1 | All-cause mortality trends: Washington D.C. and synthetic control city. The blue dotted lines represent the generalized synthetic control prediction (synthetic city) of mortality rates while the solid black line is the observed mortality rate of the Medicaid expansion city. The vertical axis of the graphs represents the per 100,000-person mortality rates. The horizontal axis of the graphs represents time units (i.e., the first month of the year). The darker gray graph areas correlate with the start and duration of Medicaid expansion in the respective city (applicable for all the figures).

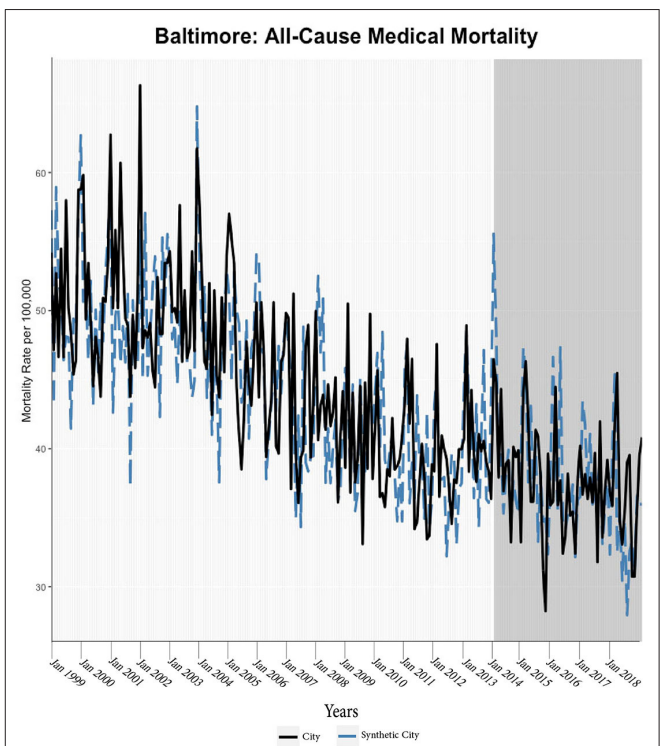

FIGURE 2 | All-cause mortality trends: Baltimore and synthetic control city.

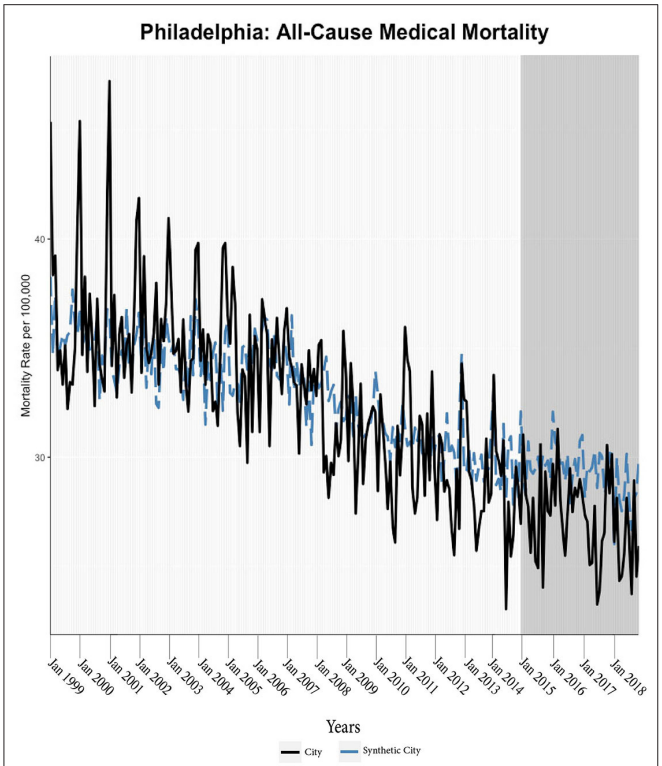

FIGURE 3 | All-cause mortality trends: Philadelphia and synthetic control city.

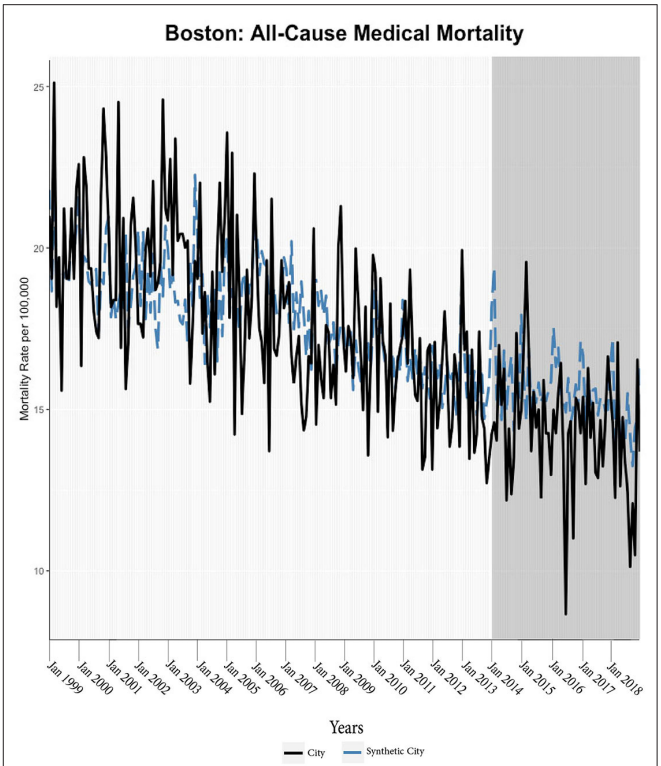

FIGURE 5 | All-cause mortality trends: Boston and synthetic control city.

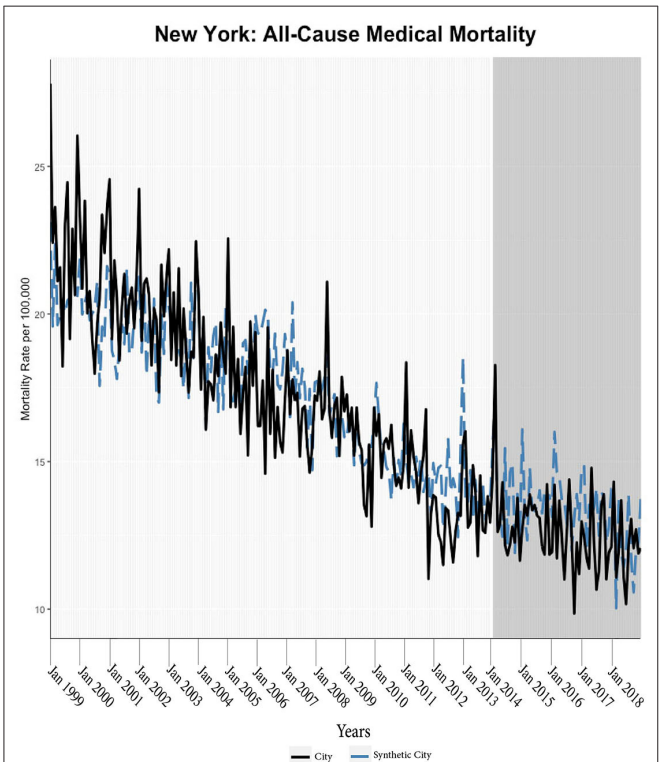

FIGURE 4 | All-cause mortality trends: New York and synthetic control city.

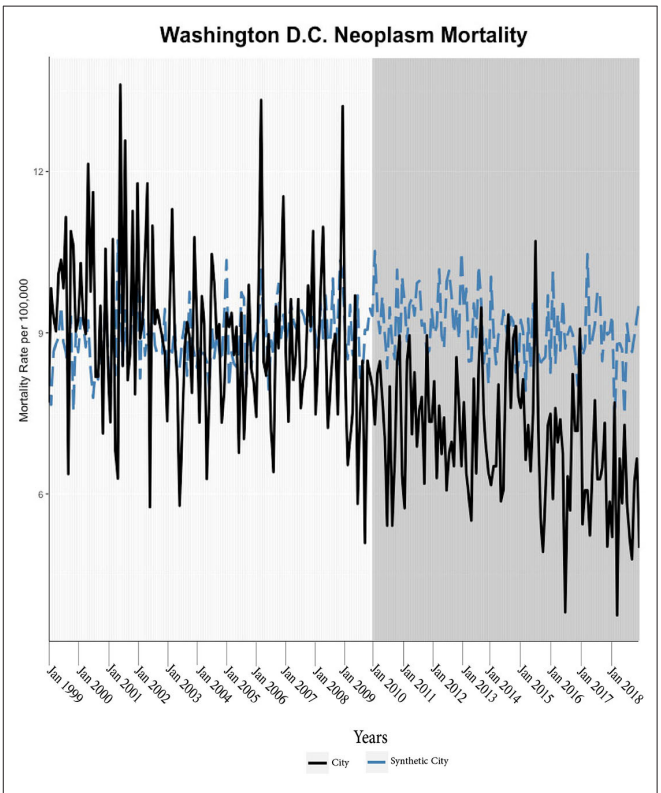

FIGURE 6 | Neoplasm mortality trends: Washington D.C. and synthetic control city.

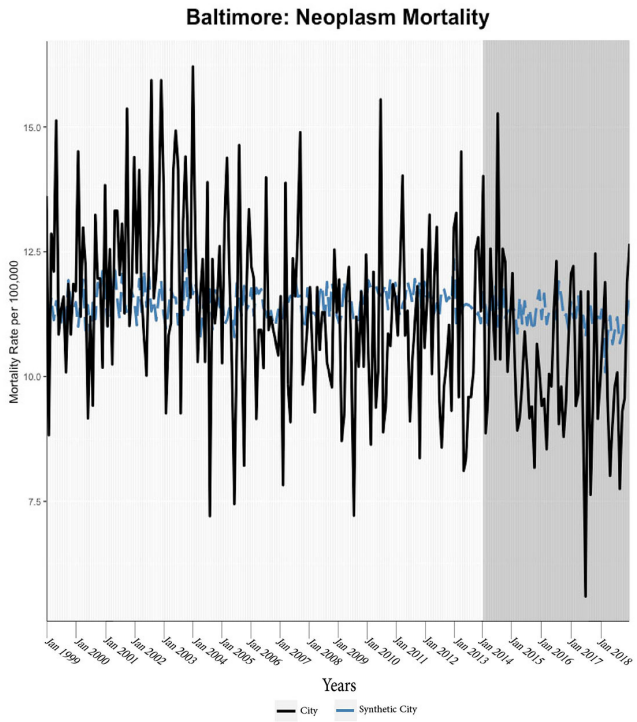

FIGURE 7 | Neoplasm mortality trends: Baltimore and synthetic control city.

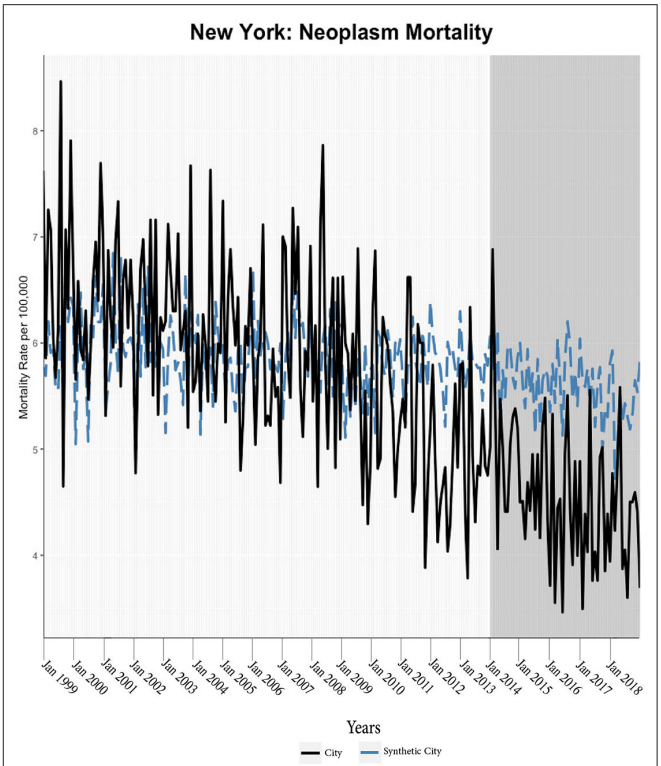

FIGURE 9 | Neoplasm mortality trends: New York and synthetic control city.

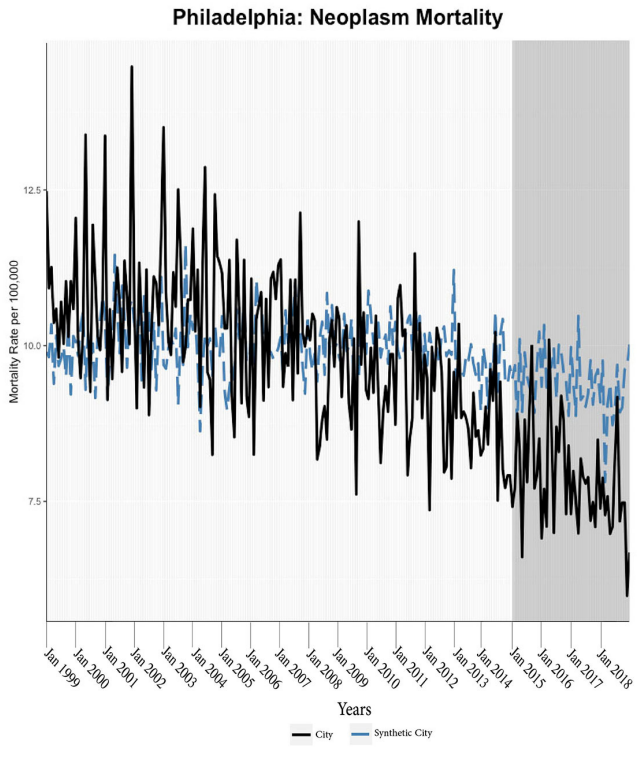

FIGURE 8 | Neoplasm mortality trends: Philadelphia and synthetic control city.

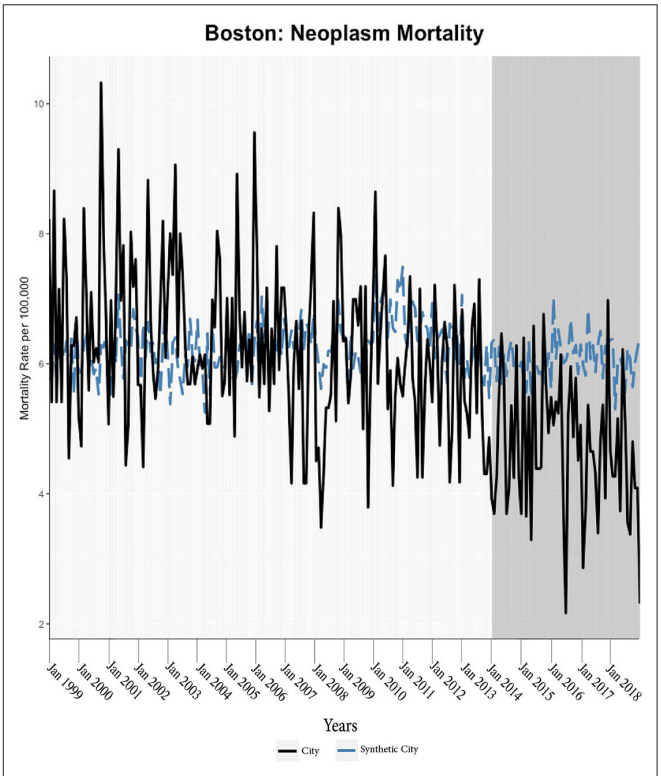

FIGURE 10 | Neoplasm mortality trends: Boston and synthetic control city.

effect sizes are seen across all cities (ATT range = −1.39 to −2.78) apart from Washington D.C., which saw a much larger decrease (ATT = −5.40). The −5.40 ATT value for Washington D.C. conveys that the average monthly neoplasm mortality rate per 100,000 was reduced by 5.40 over the post-intervention period. When assessing neoplasms, we observe a significant reduction in mortality in four cities excluding New York (**Figures 5–10**). Pooled across all cities, there was a significant reduction in the neoplasm (Population-Adjusted ATT = −1.37 [95% CI −2.73, −0.42]) and all-cause medical (Population-Adjusted ATT = −2.57 [95% CI −8.46, −0.58]) mortality rates but not cardiovascular (Population-Adjusted ATT = −3.79 [95% CI −24.57, 11.68]) and respiratory mortality (Population-Adjusted ATT = −0.91 [95% CI −6.99, 4.89]) (see Figures 12–22 in the **Appendix**).

According to established practices, we discard models that have grossly mismatched pre-intervention mortality trends between the treatment and synthetic units (27); this was only observed for respiratory mortality in Baltimore, which was highly volatile. Subsequently, the treatment effect was calculated through the ATT and CTT parameters; *p*-values indicated levels of statistical significance (**Table 2**; **Figure 11**). Parameters (ATT, %ATT, or CTT), time unit of measurement (yearly or monthly), and post-intervention period length must be considered when comparing across the reported values. Overall, our synthetic control models indicate a trend of decreases in neoplasm and all-cause medical mortality rates in cities that expanded Medicaid compared to their estimated counterfactuals. Sensitivity analyses of monthly data converted to yearly data indicated that the analysis was not sensitive to yearly data. Specifically, the conversion of monthly all-cause medical and neoplasm mortality into yearly aggregates changed significant differences into insignificant differences (see Tables 1, 2 in the **Appendix**). The in-time placebo sensitivity analysis indicated that the false intervention time point did not result in significant change in mortality rates (see Table 3 and Figures 11–20 in the **Appendix**). The weighted composition of each synthetic control model in the mortality analyses is included (see Table 4 in **Appendix**).

The generalized synthetic control analyses of insurance rates indicated that the observed city after Medicaid expansion were all much greater than predicted (**Table 3**) (see Figures 21–25 in **Appendix**). Most notably, the gains in Washington D.C. (ATT = 4.23%, $p = 0.050$), Baltimore (ATT = 3.23%, $p = 0.048$), and Philadelphia (ATT = 4.30% $p = 0.046$) were significantly greater than the predicted trends in the absence of Medicaid expansion. New York and Boston, two cities with already had high rates of coverage prior to Medicaid expansion and the derived treatment effects did not significantly differ from the predicted gains.

DISCUSSION AND CONCLUSION

Three principal conclusions can be drawn from our analysis. First, we document significant declines in all-cause medical mortality, partly driven by concurrent and significant reductions in neoplasm mortality, in most Northeastern cities following ACA Medicaid expansion. Second, while Medicaid expansion does appear to be associated with reductions in mortality rates across the cities, we observe evidence of varied effects. Third, analyses of yearly data in this study generally lacked sufficient statistical power.

Most cities experienced significant decreases in neoplasm mortality and all-cause medical mortality after Medicaid expansion as compared to the predicted rates. The potential mechanisms of this observed decline may be several. An intended function of Medicaid expansion was to decrease the uninsured rate (15, 16). While there was uptake of Medicaid coverage, much of this enrollment was moderate followed by a mild and steady increase (11). While the magnitude of increased insurance coverage may appear minor, more ill and vulnerable populations (e.g., women, African Americans, Hispanics, immigrants) gained coverage through Medicaid expansion which may explain the larger decrease in mortality observed in this study (11, 28).

New enrollees encountered reduced financial barriers which led to receiving timelier clinical and surgical care along with better ability to access prescription medicines for acute and chronic conditions (29–33). Improved self-reported and objective measures of quality as a result of Medicaid expansion accompanied increasing use of preventive care (34). In totality, Medicaid expansion reduced financial barriers and improved integration of communities in medical systems which resulted in individuals receiving more necessary and high-quality medical care (34). Additionally, Medicaid expansion has been associated with increased overall financial stability (35, 36). Greater financial stability, especially for those managing chronic conditions or suffering catastrophic events, not only allows for appropriate seeking of care but also may lead to broader health benefits. For example, families with greater financial stability are more able to achieve higher standards of living which act through material and psychosocial mechanisms to produce superior wellbeing (12, 37).

Considering neoplasm mortality, long standing research has generally considered the inability to access quality medical care as the major determinant of mortality especially in populations with lower socioeconomic status (37). Numerable studies of those with newfound access to Medicaid indicated that these individuals benefited greatly from expansion in the context of cancer care and mortality (38–42). As disease stage influences neoplasm prognosis, access to care is naturally a significant influence on neoplasm mortality (37). The longitudinal nature of oncological disease would suggest that reductions in mortality should become more profound as time passes from Medicaid expansion. Nevertheless, more proximal effects on cancer mortality due to the newfound ability to access diagnostic care and treatment have been reproduced not only in this study but also in others. Those with undiagnosed cancers were found to receive earlier screenings and diagnostic tests leading to appropriate oncological intervention (38, 42). Examining data as early as three-years after Medicaid expansion, Lin et al. found a reduction of late-stage lung cancer diagnoses and an increase in early-stage lung cancer diagnoses (41). Lung cancer as well as other aggressive forms of cancer may progress rapidly

TABLE 2 | Treatment effects on mortality rates in treated cities.

	ATT	Lower 95% CI	Higher 95% CI	ATT%[†]	ATT% Lower 95% CI	ATT% Higher 95% CI	*p*
Washington D.C. | 2010							
Circulatory mortality	−14.83	−45.20	7.47	−13.62	−41.51	6.86	0.168
Respiratory mortality	2.31	−6.21	9.73	18.39	−49.41	77.43	0.658
Neoplasm mortality	−1.95	−3.04	−0.98	−21.88	−34.10	−10.99	0.002***
All-cause medical mortality	−5.40	−12.50	−3.34	−18.84	−43.64	−11.67	< 0.001***
Baltimore | 2014							
Circulatory mortality	−7.00	−24.83	3.13	−4.35	−15.43	1.95	0.148
Respiratory mortality	1.96	−2.99	6.39	6.78	−10.33	22.08	0.482
Neoplasm mortality	−0.96	−1.90	−0.17	−8.56	−17.05	−1.53	0.01***
All-cause medical mortality	−1.76	−6.51	−0.28	−4.48	−16.53	−0.70	0.038**
Philadelphia | 2015							
Circulatory mortality	0.96	−13.22	9.47	0.86	−11.89	8.51	0.934
Respiratory mortality	−1.37	−5.10	1.43	−5.95	−22.21	6.22	0.336
Neoplasm mortality	−1.63	−2.73	−0.99	−17.17	−28.72	−10.39	< 0.001***
All-cause medical mortality	−1.97	−5.92	−0.50	−6.77	−20.29	−1.71	0.018**
New York | 2014							
Circulatory mortality	−0.39	−25.30	19.57	−0.87	−56.70	43.86	0.802
Respiratory mortality	−3.00	−10.44	4.42	−26.77	−93.16	39.48	0.322
Neoplasm mortality	−1.03	−2.86	0.203	−34.46	−95.69	6.77	0.096*
All-cause medical mortality	−2.78	−10.65	−0.13	−18.11	−69.29	−0.86	0.04**
Boston | 2014							
Circulatory mortality	−7.33	−18.72	2.50	−15	−38.31	5.12	0.144
Respiratory mortality	−0.67	−5.19	4.68	−6.07	−47.09	42.46	0.836
Neoplasm mortality	−1.35	−2.49	−0.55	−22.17	−40.93	9.06	0.004***
All-cause medical mortality	−1.39	−4.53	0.09	−8.85	−28.77	0.57	0.068*

** ≤0.10 ** ≤ 0.05 *** ≤ 0.01.*

[†] Percentage change of post-intervention mortality of Medicaid expansion city from synthetic control prediction.

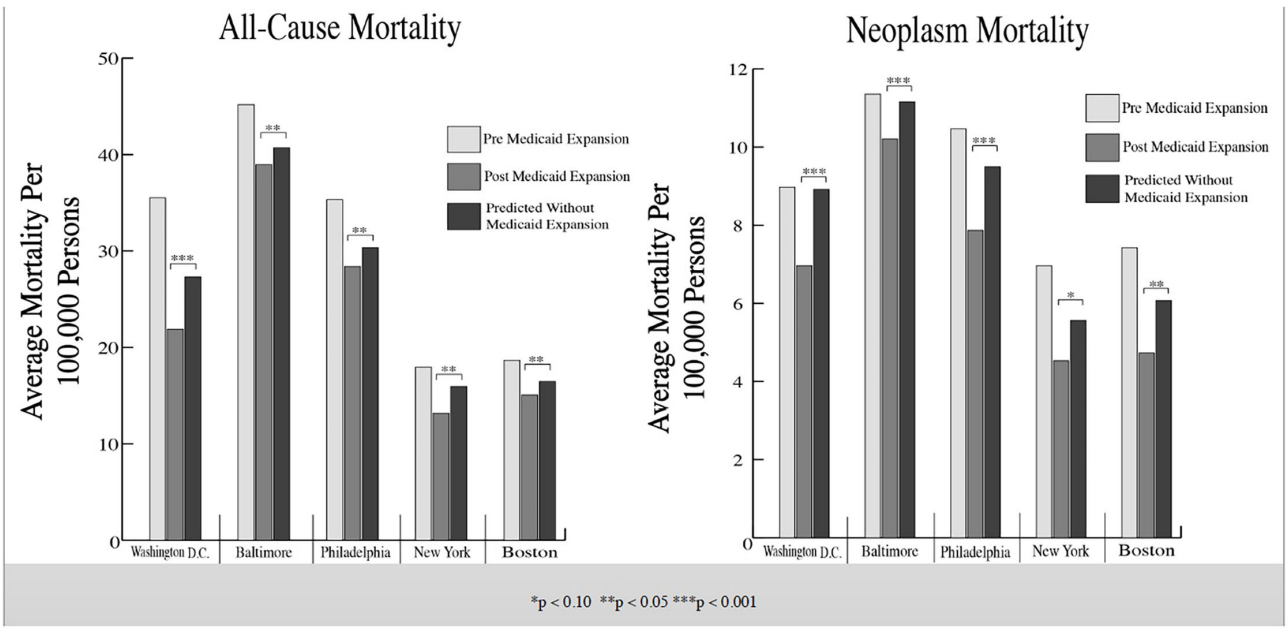

FIGURE 11 | Average mortality rates among treated cities and synthetic control cities. These bar graphs represent the average mortality rate of the selected Northeastern cities. Mortality rates before and after Medicaid expansion are illustrated along with the predicted average mortality rates if Medicaid expansion had not occurred in said cities (derived by the generalized synthetic control model).

TABLE 3 | Treatment effects on percentage of population insured in treated cities.

City | Intervention year	ATT	Lower 95% CI	Higher 95% CI	*p*
Washington D.C. | 2010	4.22%	0.28%	10.58%	0.048**
Baltimore | 2014	3.23%	0.30%	10.56%	0.046**
Philadelphia | 2015	4.31 %	0.24%	6.66%	0.036**
New York | 2014	0.86%	−13.13%	9.75%	0.860
Boston | 2014	−12.54%	−16.91%	7.80%	0.618

** ≤ 0.05.

leading to appreciable mortality that can be avoided by early diagnosis as potentially related to Medicaid expansion (41).

Neoplasm mortality reductions partly, but not fully, underlie all-cause medical mortality reductions; unrelated reductions may result from other causes that are amenable to increased access; these may or may not have been captured separately in our study.

Variations in the observed effect sizes between cities may be related to distinct policy landscapes before Medicaid expansion. Boston, for example, passed the MHCR in 2006 and this already offered coverage to many near FLP. Indeed, baseline mortality rates in Boston were lower than in other cities in our study prior to Medicaid expansion. Further, extensive pre-Medicaid expansion insurance coverage and limited baseline mortality suggest that ACA Medicaid expansion impacts be minimal in Boston. Similarly, New York City had implemented state-wide low-income coverage policies for non-elderly adults prior to the formal expansion of Medicaid in 2014 (43). Specifically, the 2001 Family Health Plus program was expanded to 100% of FLP (43); this standing program may have reduced the impact of Medicaid expansion in New York City. Such characterization of these two policy landscapes is bolstered by the results of the insurance rate analysis. Specifically, New York and Boston did not observe significant gains in the insured population as compared to their predicted trajectory absent Medicaid expansion. Nevertheless, we do observe smaller and statistically insignificant gains in the insured rate after Medicaid Expansion; given the already high rates, we anticipate that only minor gains in insurance rates would have been possible, so that this limited magnitude may not achieve statistical significance.

By contrast, the remaining cities resided in states where Medicaid expansion resulted in a large expansion in their public insurance eligibility criteria. Before its early expansion of Medicaid in 2010, Washington D.C. had only adopted narrow programs beyond the federal Medicaid requirements. Washington D.C. not only expanded Medicaid early, but the district set eligibility criteria to 210% FPL, which makes this expansion the largest of any included city (44). Similarly, Pennsylvania had heavily utilized waivers to create several targeted programs but none of these programs were broadly applicable to Medicaid expansion-eligible populations. Philadelphia had similar mortality rates to Washington D.C. but the extent of Medicaid expansion was less significant (i.e., 138% FPL).

Unlike Washington D.C. and Philadelphia, the 2006 Primary Adult Care (PAC) program under the HealthChoice program in Baltimore expanded coverage (prescription, primary care, behavioral health) to childless adults at or below 116% FPL (45). Accordingly, Medicaid expansion did moderately expand Baltimore's coverage criteria from baseline. Nevertheless, Washington D.C. and Baltimore had the highest mortality rates before Medicaid expansion; this inclines both cities to larger observed effects. Medicaid expansion operated in the context of Baltimore's elevated mortality rates, higher poverty rates, and large minority populations; these factors likely promote greater Medicaid expansion effects. The aforementioned factors suggest that Medicaid expansion might be most efficacious in Washington D.C., Baltimore and Philadelphia. The insurance rate models found significant increases in the insured percentage of said cities as compared to those trends predicted in the absence of Medicaid expansion. This provides evidence that Medicaid expansion may have increased insurance rates significantly.

Importantly, our findings diverge from state-wide analyses, in that, the magnitude of Medicaid expansion impact was larger in urban areas than in states as a whole. We attribute this to multiple factors. First, the concentration of health issues in urban areas may be more able to capture the health effects of Medicaid expansion (21). Second, greater proximity to medical care in urban environments, unlike rural areas, suggests that financial access is a stronger variable in determining healthcare access (46). With respect to Medicaid expansion specifically, previous studies have shown that medical utilization starkly increased in states after the introduction of Medicaid expansion (28). Such observed increases in utilization along with broader increases in physician supply and particularly high densities of healthcare resources in large cities strongly suggest that the increased access to care was achieved through Medicaid coverage (28, 46). This further suggests that lesser gains in urban coverage, as compared to rural populations, can be potent in improving population health in urban settings (47).

Certain limitations should be considered. Sensitivity analyses indicated that yearly data were unable to derive similarly statistically significant treatment effects as compared to monthly data. This may be explained by differences in granularity reducing study power, seasonal trends, etc. Thus, it is likely that the yearly circulatory and respiratory mortality data used are inadequate to capture treatment effects. Otherwise, mortality is an extreme marker of population health not fully representative of the total effects of Medicaid expansion. Considering trend volatility, dissimilar baseline mortality, different intervention sizes, and the fundamental limitations in the GSC, lack of findings should not be considered as precluding effects on certain types of mortality (e.g., respiratory).

Like most other Medicaid expansion studies, this study fundamentally compares mortality rates between non-ME and Medicaid expansion states where these groups may experience unique confounding influences given the ecological nature of the data. For instance, city specific events related

to police brutality, weather events, civil unrest, etc., may be lay the ground for specific forms of medical-related mortality *via* more diffuse factors (e.g., stress, slow medical emergency response times). Nevertheless, these effects may not strongly affect the sum magnitude of the observed mortality and would likely only dampen observed degree effects attributed to Medicaid expansion in the present study. The use of only urban populations, numerous independent relevant covariates, and the generalized synthetic control method allow this study to limit the influence of observed and unobserved confounding unlike most previous studies of Medicaid expansion. Regardless, overfitting is a concern in synthetic control models and while the GSC model does improve upon overfitting issues found in the traditional synthetic control methods *via* the use of semiparametric estimation and cross-validation schema, the risk of overfitting remains (25).

Unlike most literature, the county-level nature of this analysis limits the number of individuals available for study. As such, separate analyses of older sub-populations were inviable given the limited size of such a sub-population and the risk of stochastic variability biasing the model. Nevertheless, the inclusion of younger and healthier populations in the presented results more likely underestimates the treatment effects of Medicaid expansion (28). Similarly, the nature of the data utilized county-wide covariates, as such analyses of mortality rates in subsets of particular covariate categories (e.g., education-level) was unable to be conducted. Further studies are required to determine whether the observed effects of Medicaid expansion can be generalized to other cities without Medicaid expansion. To this point, each city's results must be considered given the unique treatment intensity and policy history characterized above.

Significant reductions in multiple forms of urban mortality were attributed to Medicaid expansion. The degree of effects was seemingly related to baseline mortality rates, prior expansion status, and the magnitude of Medicaid expansion. Our study indicates that Medicaid expansion saved lives in the included urban settings.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

CA designed and conducted the initial gathering of data, organization, analysis, and organized the findings and subsequently put the information in a manuscript format. PP was involved in the data analysis, figure making, and the writing of the manuscript. SA was involved in the organization and the writing of the manuscript. All authors contributed to the article and approved the submitted version.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.707907/full#supplementary-material

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frontiers in Public Health

ORIGINAL RESEARCH
published: 13 January 2022
doi: 10.3389/fpubh.2021.793973

Requirements for Creating a Position for Community Health Nursing Within the Iranian Primary Health Care System: A SWOT Analysis

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Background: Accepting community health nursing in the primary care system of each country and focusing on creating a position for community health nurses is of significant importance. The aim of this study was to examine the stakeholders' perception of the requirements for establishing a position for community health nursing in the Iranian primary health care system.

Methods: This qualitative study was done using 24 semi-structured interviews conducted from May 2020 to February 2021 in Iran. The participants were selected through purposive sampling and consisted of nursing policy makers, the policy makers of the Health Deputy of Ministry of Health, the managers and the authorities of universities of medical sciences all across the country, community health nursing faculty members, and community health nurses working in health care centers. After recording and transcribing the data, data analysis was performed in MAXQDA10 software, using Elo and Kyngas's directed content analysis approach and based on WHO's community health nursing role enhancement model. The statements for each main category were summarized in SWOT classification. To examine the trustworthiness of the data, Lincoln & Guba's criteria were used.

Results: By analyzing the interviews 6 main categories identified consist of creating a transparent framework for community health nursing practice, enhancing community health nursing education and training for practice in the primary health care system and community settings, seeking support, strengthening the cooperation and engagement among the key stakeholders of the primary health care system, changing the policies and the structure of the health system, and focusing on the deficiencies of the health system. Each main categories including the subcategories strengths, weaknesses, opportunities and threats (SWOT).

Conclusions: Based on the participants' opinions, focusing on the aforementioned dimensions is one of the requirements of developing a position for community health

OPEN ACCESS

Edited by:
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Reviewed by:
Katalin Dr. Papp, University of Debrecen, Hungary
Birsen Altay, Ondokuz Mayıs University, Turkey

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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *12 October 2021*
Accepted: *09 December 2021*
Published: *13 January 2022*

Citation:
Hosseinnejad A, Rassouli M, Jahani S, Elahi N and Molavynejad S (2022) Requirements for Creating a Position for Community Health Nursing Within the Iranian Primary Health Care System: A SWOT Analysis. Front. Public Health 9:793973. doi: 10.3389/fpubh.2021.793973

nursing within the Iranian PHC system. It seems that correct and proper implementation of these strategies in regard with the cultural context of society can help policymakers manage challenges that prevent the performance of community health nursing in the health system.

Keywords: community health nursing, primary health care, SWOT analysis, Iran, policy

INTRODUCTION

The health system all around the world, including Iran, has undergone many changes during the last 20 years (1, 2) and will have encountered serious challenges by the year 2050 (3). Lifestyle changes will increase life expectancy, followed by an increase in the incidence and the prevalence of non-communicable diseases (4–8). The emergence of new diseases and pandemic crises such as COVID-19 will pose new challenges to the health systems of different countries (9, 10). On the other hand, adopting approaches to reduce the length of hospital stay, increasing outpatient surgeries, focusing on health promotion, and the desire to reduce costs will lead to a shift in paradigms, transferring health care provision from hospitals to community settings (11–13). The emerging challenges will focus on the need to change, enhance, or reform the health system (14, 15). Due to the important role of family and community in disease management and health promotion, health systems around the world will change into community-based nursing care (5, 16). Since community health nurses play an important role in supporting the transfer of hospital care to home and health centers such as comprehensive health centers, in order to address the community needs and provide cost-effective care, community health nursing services should be used (17, 18).

Community health nurses play a key role in providing quality primary health care and universal health coverage (19, 20). Nurses can take leadership roles and provide direct health care based on patient, family and health system's priorities in primary care. Care seekers are also satisfied with the services provided by community health nurses (21). Community health nurses take roles in the 4 areas of primary health care including *preventive care* (health promotion, education), *management of chronic diseases* (coordination of care, control and assessment, rehabilitation), *practical actions* (the health of children and the elderly, midwifery care), *involvement in health policymaking* (developing policies, planning, evaluation and supporting national programs in accordance with the situations, requirements and priorities) (22).

Community health nursing has grown significantly in the health system of all developed countries around the world (23). According to the WHO, more than 42% of community health nurses in the United States work in health centers. Given the fact that many actions in primary health care do not require a physician's knowledge or skills, the tendency to use community health nurses for developing the capacity of primary health care is on the rise. Replacing a physician with a nurse is a strategy that will improve the accessibility, effectiveness, and quality of health care (17, 24). In some European countries, physician-based and hospital-based approaches are replaced with community health nurses and these nurses provide health care and services to the members of the community (25, 26). Among European countries, Finland has the lowest rate of physician-patient contact, and a large number of community health nurses provide services in the health system instead of physicians. In Ireland, based on an efficient primary health care model, more integrated services at all levels of prevention, especially the first level, are provided by community health nurses to people of all ages from infants to the elderly. There are also community-level clinics directed by community nurses for providing care and prescribing medication (27). In the United Kingdom, a model exists for involving community health nurses in primary health care, in the form of clinic management, the provision of health care, education and counseling services, and even the competence to prescribe medication (15).

In Iran, the courses of community health nursing and epidemiology were included in the undergraduate curriculum after community-based and community-oriented nursing disciplines were developed in 1985 by policymakers. However, community health nurses currently encounter obstacles and challenges in offering specialized services in the form of specialty job descriptions developed by the Ministry of Health (28, 29). Currently, comprehensive urban health centers are run by graduates and associates in family health, environmental health, occupational health, disease control, and midwives and local health centers, by Behvarzes (rural health care workers), in order to provide health services. In these centers, health services are provided sporadically (28) and no effective strategies in line with community needs are implemented for care provision (30, 31). On the other hand, neither following up discharged patients and vulnerable groups such as chronic patients, the elderly, pregnant women, and infants at the community level, nor offering home visits with the aim of promoting health is done at comprehensive health centers. Despite the fact that home care services are one of the most important components of the health system, they are not yet institutionalized within the structure of the primary health care system (5). Furthermore, for addressing the challenges of the health system stated in the 6th Development Plan, the Iranian Parliament urged the Ministry of Health to provide a *comprehensive health services system* by prioritizing health and prevention over treatment and to offer primary health care by focusing on the referral system, family physician program, and the provision of home-based and community-based nursing care. The most important action taken by the Nursing Deputy of the Ministry of Health for addressing

challenges against the establishment and development of home-care and nursing consultation centers was the formulation of regulations for hospices and long-term care centers (4).

Community health nurses use comprehensive approaches, as well as clinical and managerial knowledge and skills, to lead and manage care and coordinate the continuum of care transfer from hospital to home and the community. They also have the competence to coordinate the care team and interdisciplinary participation, manage patient transfers from hospitals and medical centers to comprehensive health centers, and perform their follow-up at the community level. Therefore, these services can be assigned to them (32). In terms of cost-effectiveness, various studies show that providing community-based care by community health nurses instead of hospital care leads to improved health and quality of care, chronic disease management, patients' access to community-based social services, and patient satisfaction, which in turn results in reduced ER visits and hospitalization, eventually decreasing treatment costs. In several US states, including Michigan, the community-based care delivery model has been implemented resulting in reduced overall health costs (18, 33).

Considering the effectiveness of analyzing the stakeholders' perceptions in policy-making, management and development of the health system (34), it is important to examine their views to investigate the weaknesses, strengths, opportunities and threats of community health nursing in order to identify the necessary measures for reforming the primary health care system, which is why SWOT model was used in the present study. Models can help researchers with identifying, describing, explaining or predicting (35). SWOT is a simple conceptual framework that can be used by individuals, groups, teams, and organizations providing health care (36). The aim of this study was to explain the stakeholders' perception of the requirements for establishing a community health nursing position within the Iranian primary health care system.

METHODS

Study Design

This is a qualitative study using directed content analysis.

Study Settings and Participants

Twenty-four subjects participated in this study. The researcher first interviewed the key informants, namely nursing policymakers, the policymakers affiliated to the Health Deputy of the Ministry of Health, the health managers and the authorities of the Iranian universities of medical sciences. Then interviews were conducted with other participants, including community health nursing faculty members and community health nurses working in health centers and various provincial deputies. The participants were selected through purposive sampling. The inclusion criteria for the participants consisted of having experience in policymaking and decision-making in the field of health and community health nursing, and voluntarily participation in the research. The time and the place of the interviews were fixed according to the participants, at their own workplaces and offices. It should be noted that due to COVID-19 pandemic conditions, some interviews were conducted via Skype or Whatsapp.

Data Collection

The research data was collected using in-depth semi-structured interviews conducted from May 2020 to February 2021. Each interview took 30–70 min. The interviews continued until data saturation was achieved, or in other words, until new data did not modify or further develop the model and no new classes were created (37). Prior to the interviews, the interview questions guide was developed based on the policies of WHO model (20) separately for each participant group in order to ensure the comprehensiveness of the collected data. The questions included "According to your experiences, what is the position of community health nurse in Iranian health system?," "How can we define a position for community health nurses according to their duties regarding public health promotion?," "What are the opportunities in and barriers against providing community-based nursing services in Iran?," "What are the executive strategies for nurses to enter the network system?." Furthermore, in order to collect more information during the interviews and clarify the content, exploratory questions were asked such as "Can you explain more? Can you give an example?"

Research Framework

The present study was designed and implemented based on the model "Enhancing the Role of Community Health Nursing for Universal Health Coverage" (WHO 2017). This model is a guide, a framework, and a strategy to strengthen and enhance the role of community health nursing with the aim of universal health coverage, which is considered a comprehensive action plan focusing on national and local strategies. According to the evidence obtained from a WHO study during 2010–2014, community health nursing is regarded as an important component in primary health care in 22 countries affected by a shortage of service providing manpower. In this model, four major policies were proposed to manage the challenges of community health nursing and promoting the role of community health nurses (20). The policies include *the development of a clear framework for community health nursing practice*, *promoting the education and training of community health nurses for working in the primary health care system and community settings*, *strengthening cooperation and partnership among the major stakeholders of primary health care system*, and *the development of comprehensive support plans for community health nurses in various countries*.

Data Analysis

The data was collected and analyzed simultaneously. Data management was done using MAXQDA-v10 software (38). Data analysis including preparation, organization and reporting was done as proposed by Elo & Kyngas (39, 40) (**Table 1**). An example of data analysis is shown in **Table 2**. SWOT (Strengths, Weaknesses, Opportunities, and Threats) analysis was carried out through the content analysis of interviews. The statements of each main category of the study were integrated into SWOT classes (**Table 3**). The strengths and weaknesses are the internal

TABLE 1 | The process of qualitative data analysis based on Elo & Kyngas' Method.

Data preparation	Selecting the unit of analysis	After turning the interviews into texts, the manifest content (such as interview text) and latent content (non-verbal behavior of participants) were analyzed and semantic units were identified.
	Finding a logical connection between data and the topic in general	The interview texts were reviewed several times by the researcher, and the researcher was constantly engaged with data for a long time until data saturation was achieved.
Data organization	Creating an analysis matrix	At this stage, a unconstrained matrix was created and the following were extracted as main categories: (1) developing a transparent framework for community health nursing practice, (2) enhancing community health nursing education and training for practice in the primary health care system and community settings, (3) strengthening cooperation and engagement among the major stakeholders of the primary health care system, and (4) the development of comprehensive support programs for community health nurses.
	Data extraction from content based on classes	The possibility of placing generic categories in the main categories of the matrix or the formation of new categories was investigated, based on conceptual and logical connections.
	Categorization	The number of codes decreased by the merging of similar codes into more general codes, consequently forming the generic categories.
	Classification	The created categories were classified based on the similarities and differences and similar categories were merged.
	Abstraction	The categories discovered in the initial main categories were placed into the analysis matrix. In case of inconsistency with the existing classes, new main categories were created by merging similar classes (new classes in the current study: *changing the policies and the structure of the health system* and *focusing on the deficiencies of the health system*)
Reporting		The sampling process, the participants' characteristics, data collection, data analysis, and the analysis of each class are thoroughly reported under *Findings*.

TABLE 2 | An example of data analysis.

Main category	Generic categories	Subcategories	Primary codes	Quotation
promoting the education and training of community health nurses for working in the primary health care system and community settings	Modifying the undergraduate education system	The necessity of nursing education system's being community-based	- The individual-based undergraduate education system - Disease-based approach in the education - Not being prepared for community-based services	"Practically, the undergraduate education system is not community-based, but is basically individual-centered, and even patient-centered, which still focuses on diseases. Not much attention is paid to the subject *individual care seeker* and the main focus is on the disease-based approach. When you teach someone in accordance with such a system, in my opinion, one is not prepared to offer community-based services."

aspects of the professional position of community health nursing in the Iranian primary health care, related to nursing schools and the nursing system, while the opportunities and threats include external environmental factors other than colleges and nursing system. An example of SWOT analysis results is displayed in **Table 4**.

Rigor and Trustworthiness

In order to enhance the rigor and the trustworthiness of the data, the four indicators credibility, dependability, confirmability, and transferability of Lincoln and Guba were used (41). Data credibility was determined through long interaction with data, member check, peer review, and reviewing the interview texts by the participants. Data dependability was ensured through responsiveness. In other words, the process of research and data analysis was examined by an external supervisor familiar with qualitative research. In order to ensure confirmability, all the steps of research are presented and explained in detail. Data transferability was achieved by providing a deep and rich explanation of the findings and the maximum amount of sample variance.

Ethical Considerations

This study was approved by the Ethics Committee of Ahvaz Jundishapur University of Medical Sciences (IR.AJUMS.REC.1398.874). Ethical considerations in this study included the voluntary nature of research participation, the explanation of research objectives to the participants, and ensuring their anonymity, their right to withdraw from the study, and the confidentiality of the data.

TABLE 3 | A summary of the statements of the SWOT analysis based on 4 subcategories.

Main category		
Creating a transparent framework for community health nursing practice	**Strengths**	**Weaknesses**
	Community health nurse's ability to manage and follow up on chronic patients with regard to the previous experiences of the holistic view of community health nursing	Not recruiting community health nurses for supplying community health services
	Opportunities	**Threats**
	Redefining the duties of comprehensive health centers in the development plan, and assigning a health care title for nurses	Considering a therapeutic role for nurses working in health sectors and health managers' unfamiliarity with nurses' capabilities in the field of health The lack of need for developing a position for community health nurses in the primary health care system from the perspective of the senior managers of the Ministry of Health
Enhancing community health nursing education and training for practice in the primary health care system and community settings	**Strengths**	**Weaknesses**
	Thorough and comprehensive theoretical framework for the community health nursing graduate program	Deficiencies in offering practical courses in undergraduate and post-graduate community health education programs Deficiency in practical courses due to inappropriate fields Low quantity and quality of community health nursing instructors Poor community-based and community-oriented nursing education Community health nursing curriculum's not being tailored to community health needs
	Opportunities	**Threats**
	Offering a previously-developed design to present post-graduate courses for nurses' employment in health centers Offering specialty training to community health nurses in undergraduate programs	Considering a therapeutic role for clinical disciplines such as nursing by the Ministry of Health
Seeking Support	**Strengths**	**Weaknesses**
	The existence of nursing boards and associations effective in seeking support	Community health nursing leaders' not taking actions to introduce this discipline and not publicizing information in this regard Lack of interactions with the policymakers of the Ministry with the aim of presenting community health nurses' capabilities Insufficient measures taken by the nursing system organization in support of community-based nursing
	Opportunities	**Threats**
	The existence of a position and deputy for nursing (Nursing Deputy)	Public unawareness of community health nursing and the capabilities of community health nurses Highlighting the therapeutic role of nurses in the media
Strengthening the cooperation and engagement among key stakeholders of the primary health care system and	**Strengths**	**Weaknesses**
	-	Separate actions taken by various departments in universities of medical sciences The lack of common courses between Health and Nursing departments in post-graduate education
	Opportunities	**Threats**
	Nursing Deputy's power and capacity in establishing communication with other deputies of the Ministry, especially the Health Deputy	The lack of interdisciplinary interaction between health disciplines Tribalism in the health system
Changing the policies and structure of the health system	**Strengths**	**Weaknesses**
	Community health nurses' interest in working in health sector	Not being motivated to work in the Family Physician Plan and health sector due to insufficient salaries and payments The lack of an effective nursing leadership across the country Nursing managers' taking no actions in order to extend the boundaries of nursing practice The shortage of nursing clinical workforce Barriers against employing nurses in the health sector Low enrollment capacity in community health post-graduate programs
	Opportunities	**Threats**

(Continued)

TABLE 3 | Continued

Main category		
	The existence of job titles for nurses and health care giver in the PHC Operational objectives of Nursing Deputy regarding holistic nursing The focus of senior managers of the health system on legalization of nursing services at the community in the form of home-care and consultation centers	Not giving health priority over treatment in the health system and focusing on the secondary prevention Conflicts of interest with other medical disciplines The lack of power to change and modify the network system Insufficient funding and budgets for employing community health nurses
Focusing on the deficiencies of the health system	**Strengths**	**Weaknesses**
	Better nursing responsiveness to the public's health needs Empowering nurses for the coordination of medical and care plans	High workload of caregivers and low manpower-population ratio Routinized activities of comprehensive health centers and ignoring the personal needs of care seekers The lack of public trust in care and consultation offered by health caregivers regarding chronic diseases
	Opportunities	**Threats**
	The possibility for health system policymakers to use nursing care plan at home in order to employ community health nurses	Deficiency in the service delivery system and the referral system and not covering all areas of prevention Entrusting health policymaking management to a conscripted physician who is not familiar with people in comprehensive health centers health center physicians' focusing on therapeutic measures instead of providing health promotion services Behvarzes' not addressing people's health needs

RESULTS

Twenty-four subjects participated in this research, including 6 nursing policy makers and policy makers of the Health Deputy of Ministry of Health, 7 health managers and authorities of universities of medical sciences, 8 community health nursing faculty members, and 3 community health nursing working in health centers. The mean age and working experience of the participants were 53.5 ± 9.83 and 24.91 ± 6.09 years, respectively. 54.2% of the participants were male, and 45.8%, female. The inclusion criteria for community health nursing faculty members consisted of having at least 10 years of working experience in educational environments. The inclusion criteria for policy makers consisted of managerial experience in the health system in the field of health or nursing. Community health nurses had to have at least 5 years of working experience in health centers.

After continuous analysis and comparison, 780 codes were extracted, and 76 subcategories, 18 generic categories, and 6 main categories were identified. The main categories included *creating a transparent framework for community health nursing practice*, *enhancing community health nursing education and training for practice in the primary health care system and community settings*, *seeking support*, *strengthening the cooperation and engagement among the key stakeholders of the primary health care system*, *changing the policies and the structure of the health system,* and *focusing on the deficiencies of the health system*, each of which consisting of 4 subcategories: *strengths*, *weaknesses*, *opportunities,* and *threats*.

Creating a Transparent Framework for Community Health Nursing Practice

In order to create a transparent framework for community health nursing practice, it is necessary to make huge policies in the Ministry of Health. A transparent framework for community health nursing practice includes the explanation of a position for the community health nurse, a scope for their services, and a clear job description for them.

Strengths

The participants referred to the community health nurses' ability in managing and following up on patients with chronic diseases at the community as a strength of the health system. "In my opinion, today, we offer individual-level health care through Behvarzes and caregivers, but in order to adopt a community-oriented to the management of chronic diseases, we need a number of capable people at a higher level, such as these community health nurses who can be employed in comprehensive health centers," the director of the department of non-communicable diseases in one of the provinces said in this regard, "In old diabetes clinics, the presence of a nurse, and using teamwork was a successful experience in the management of diabetic patients."

Weakness

The participants mentioned the recruitment of community health nurses only in academic and clinical areas, the inability of nursing managers to employ community health nurses in the network system, and community health nursing graduates' not

TABLE 4 | Main categories, generic categories, and sub-categories extracted from the content-driven analysis of the interviews.

Main categories	Generic categories	Subcategories
Enhancing the community health nursing	Creating a transparent framework for community health nursing practice	Explaining the position of community health nurse
		Community health nursing service areas
		Job descriptions for community health nurses in comprehensive health centers
	Enhancing community health nursing education and training for practice in the primary health care system and community settings	Reforming the undergraduate education system
		Reforming the community health nursing post-graduate program
		Flaws in the educational system input
	Seeking support	The necessity of establishing a communication channel with the public
		The necessity of receiving support from nursing policymakers
	strengthening the cooperation and engagement among key stakeholders of the primary health care system	The Key stakeholders' role
		The necessity of interdisciplinary cooperation in education
	Changing the policies and structure of health system	The barriers against the provision of community health nursing at the level of Health and Treatment Deputy
		The barriers against the provision of community health nursing at the level of Education Deputy
		The barriers against the provision of community health nursing at the community level
		The necessity of effective performance of community health nursing policymakers
		The necessity of strengthening community health nursing institution
	Focusing on health system deficiencies	Ignored services of the health system
		Deficiencies in the network system's functioning
		Deficiencies in the healthcare team's performance

taking actions to develop a position for their own discipline. "Officially, no position exists for community health nurses in the charts of comprehensive health centers, in other words, they have no place. The only position for community health nursing is acting as a faculty member in the health departments of nursing schools."

Opportunities

Considering a the nurses at the level of PHC in the health promotion plan was considered, by the participants, as an opportunity to create a framework for community health nurse practice. "All across the country, in all urban and rural centers we have nursing titles for health nursing experts in the health organizational chart," the executive director of the Health Deputy in one of the provinces said, "if you take a look at the health chart, you will realize that the nurse is regarded at the level of PHC, and nursing managers can define this position for community health nursing through consulting with the Deputy Minister of Health."

Threats

Not needing to create a position for community health nurses in the primary health care system from the perspective of the managers of the Ministry of Health is one of the most important threats to creating a framework for community health nursing practice. "Community health nursing is a concept, up to 80% of which is currently realized by Behvarzes and healthcare providers. Given the current state of the health system, there is no need to create a position for community health nurses. Such workforce should not be placed at higher levels," the Deputy Minister of Health said in this regard.

Enhancing Community Health Nursing Education and Training for Practice in the Primary Health Care System and Community Settings

Enhancing the education and training of community health nurses in accordance with the needs of the society is one of the goals of the educational system. To this end, it is necessary to reform the educational system in accordance with the needs of society and change the educational approach from being hospital-based to being community-based. It is also necessary to focus on the inputs of the educational system, which includes a sufficient number of experienced community health nursing faculty members and appropriate fields of education in community health nursing.

Strengths

The participants referred to thorough and comprehensive theoretical frameworks in community health nursing post-graduate program. "There are no theoretical issues in community health nursing post-graduate curriculum and it is even being reviewed, because it should be reviewed every five years. But the implementation of the program is very important, and it is the teacher who plays the key role in this regard," a member of the community health nursing faculty said on the matter.

Weaknesses

The participants referred to the inefficiency of practical courses in community health nursing curriculum due to improper fields and the weakness of community-oriented education in the

nursing education system as the most important weaknesses. "The environment is not suitable for the practice of health traineeship. Trainees mostly end up in health centers because the interdepartmental systems are not ready for the community health nurses home visits, which makes the trainings impractical. The only thing that works is teaching in the classroom. At best, our nurses are well-trained for clinical purposes," one of the community health nursing faculty said regarding the importance of teaching practical community health nursing courses.

Opportunities

The prticipants regarded the possibility of training community health nursing at undergraduate level as an opportunity. "It would be excellent if nursing schools accept to train some nurses, from the beginning at the undergraduate program, tailored to the characteristics of community health nursing including the provision of environmental health services, health education, nutritional health, maternal health, fertility and immunization," said the Deputy Minister of Health in this regard, "In other words, it will be good if there is a bachelor's degree program in community health nursing. If we modify the education system in this way, our nurses will no longer have many options to be able to choose from among angiography, pediatrics, surgery and health at the same time and decide on their own. In that case, we have the workforce and we can organize it."

Threats

The participants referred to the need to reform the Ministry of Health's educational perspective of clinical disciplines and argued that education should be revised with the aim of becoming community-oriented. "In general, the therapeutic perspective is dominant in the Ministry of Health, which means that the main problem lies in the education system. Treatment perspective is dominant, not health and community-oriented views. Nurses spend little time in health departments, while they spend a lot of time in hospitals as trainees, and they even interpret ECG better than a physician. But the same nurses have no knowledge when it comes to working at the community level, and cannot provide simple trainings to people," said the Deputy Minister of Health of one of the provinces on the matter.

Seeking Support

Seeking support requires the creation of a communication channel with the public in order to increase public awareness of community health nursing discipline and see support from nursing policymakers to introduce this profession to the public.

Strengths

The participants considered the existence of nursing boards and associations to be effective in supporting community health nursing.

"Seeking support requires actions taken by nursing institutions such as nursing boards and scientific nursing associations to state the capabilities of community health nurses and allow them to demonstrate their capabilities to the health system and the public. More efforts should be made in this regard and the Scientific Association of Community Health Nursing should be more active," said a member of the community health nursing faculty member on the topic.

Weaknesses

The participants referred to the need to establish a communication channel between community health nurses and the public for support, and believed that people do not know community health nursing at all. "The leaders of community health nursing do not try to introduce the discipline either at the university level or to the people of the community. However, if they define themselves as the community health care institution and the provider of public health services, people will seek health services from them. Introducing the discipline can be very effective in seeking support because once the discipline is recognized, organizations will make a recruitment call. Introduction should be done hierarchically downwards at different levels," said one of the nursing board members in this regard, "Many in the Ministry of Health are, themselves, unaware of the existence of this type of workforce with such capabilities, because everyone we trained was recruited by the clinical and academic nursing community."

Opportunities

The participants introduced the position of Nursing Deputy as one of the most effective opportunities for strengthening the community health nursing institution, and believed that it plays a significant role in the entry of nurses into the network system. "The Nursing Deputy itself is a good opportunity, where recently, PhD graduates have been working. We also have individuals whose experience can be helpful," said a community health nursing faculty member in this regard.

Threats

From the participants' perspective, the most important threat is highlighting the therapeutic role nurses in the media. "Highlighting the therapeutic aspect of nursing by the Islamic Republic of Iran Broadcasting on occasions such as the Nurses Day, has led to the public's unawareness of community health nursing and the capabilities of this discipline in providing health and preventive services," another community health nursing faculty member stated in this regard.

Strengthening the Cooperation and Engagement Among Key Stakeholders of the Primary Health Care System

Strengthening the cooperation and engagement among key stakeholders requires actions to be taken by Nursing and Health deputies, designing programs to encourage public engagement, and the interaction of community health nursing professors with the professors of health departments.

Weaknesses

The participants regarded the departments' lack of knowledge of each other at a medical university, and the lack of interdisciplinary engagement among different disciplines of the Ministry of Health as the most important weaknesses. "The most important issue in our country is that actions are taken

dispersedly. In other words, academicians in one department at a university are not aware of another department at the same university. They may not know about each other and what others are capable of. When we get acquainted with a colleague in another department, we will get to know each other's capabilities and what our fields of study have in common," a nursing board member said in this regard.

Opportunities

The participants mentioned the Nursing Deputy's capacity and capability in communication with other deputy ministries, especially the Health Deputy, with the aim of strengthening interdisciplinary cooperation. "Today, with the help of competent individuals, we expect the Nursing Deputy to take an effective step in providing health services through interaction and communication with the Health Deputy with the aim of strengthening the cooperation between community health nurses and the health staff," one of the nursing policymakers said in this regard.

Threats

The participants considered interdisciplinary interaction necessary for strengthening cooperation and engagement. "Unfortunately, in health-related disciplines, no interdisciplinary interactions exist and our country's health system is governed through trade unionism and tribalism. As long as there is unionism, we should not expect inter-professional cooperation and engagement," another nursing policymakers stated on the matter.

Changing Policies and the Structure of the Health System

It is necessary to change the policies and the structure of the health system in order to prioritize health over treatment, change the structure of the network for community health nurses' entry to the primary health care system, resolve infrastructure issues (for instance, through the approval of supportive laws for community health nurses), issue permits for the establishment of community health clinics according to the structure of the referral system, and facilitate the insurance coverage of community health nursing services.

Weaknesses

The participants considered the community health nurses' reduced motivation to work in the health departments compared to the treatment sector as a weakness, and referred to the need to reform the payment system in the health sector. "Many senior nurses are reluctant to work with us due to low salaries and financial benefits in the health sector. Currently, a nurse in the treatment sector has a monthly income of about 10 million Tomans, which is reduced to 3 or 4 million Tomans when it comes to health, a very low amount," the executive director of the health deputy of one of the provinces said in this regard, "The job that a nurse gets in a hospital is very, very different from the job that we do in the health sector. Thus, nurses prefer to work with their bachelor's degree rather than providing health services with a master's degree. Another problem is the shortage of manpower. For this reason, the Treatment Deputy recruits all nurses, even community health nursing postgraduates, and does not give them to the Health Deputy."

Strengths

The participants stated community health nurses' interest in working in the health sector as one of the strengths, and believed by providing proper infrastructure this advantage can be used to benefit from the capabilities of community health nurses.

"Many nurses entering the community health discipline liked working in the health sector because of greater independence compared to clinical jobs and were interested in working with people at the community level," one of the community health nurses working in a health center said on the subject.

Threats

From the participants' perspective, in the health care system, hospital care takes precedence over community-based services. "It is true that we call it the Ministry of Health, but we do not see the priority of health over treatment. Our system is not health-oriented, but treatment-oriented and patient-oriented, because there is a conflict of interest," another nursing policymaker said, "For instance, in Shahrak-e Gharb, there are lots of private hospitals and more are still being launched, but not even one preventive clinic at the community level can be found. This shows that we do not access preventive procedures and cannot take preventive measures, one of the requirements of which is community health nursing."

Opportunities

The goal of making nursing community-oriented was considered as an opportunity by the participants. "Fortunately, the Deputy's strategy is community-based. On the other hand, the governmental authorities and some of the ministers have legalized the provision of nursing services for patients at the community in the form of home care and consultation centers. But we believe that this position should also exist in preventive areas in the form of nursing services in clinics, health centers, and health complexes." one of the nursing policymakers said, in this regard.

Focusing on the Deficiencies of the Health System

This class considers the flaws in the functioning of the network system and the performance of the health team as the deficiencies of the health system.

Weaknesses

The participants regarded the disproportionate manpower-population ratio, and the routinized work of the staff of comprehensive health centers without considering the individual needs of the care seeker as significant weaknesses.

"Unfortunately, due to their high workload, our community health workers are not able to provide care and follow up on patients with chronic diseases like diabetes and hypertension in proper way. People do not trust health centers with the care provided for chronic diseases, because measuring blood pressure

and weighing patients is also done by our health care providers. Since people receive better services from private centers than the ones we offer, they won't come to us in health centers." said the executive director of the Health Deputy in one of the provinces.

Strengths

The better responsiveness of community health nurses to the public's health needs, according to nurses' education in regard with diseases and care compared to the midwifery and health students was seen as a strength by the participants.

"In our health centers, a community nurse according to their knowledge, can very well-fulfill their duties in health promotion, education and prevention based on the needs of the people and follow-ups of chronic patients at the community, as well as follow-up of the patients discharged from the hospital." one of the health policymakers of the Ministry of Health said in this regard, "even one of our BSc graduate nurses has the ability to know the community and has the power to establish a communication with the workforces lower in hierarchy and those higher (i.e., physicians), which is a great advantage for the nurse, making them capable of making the necessary coordination in the field of health. We can even have a community nurse provide home care, and accordingly, we have to define *community nurse* based on the population covered by that health center."

Opportunities

Participants believed that health system policymakers should use the home care nursing plan to employ community health nurses in this field and develop the profession in various areas of the community.

"Home nursing care and consultation plan was a very good plan that health system policymakers, both in nursing and health, could use to provide services to people at the community level by employing specialist nursing workforce, especially community health nurses and even nursing experts. But unfortunately, this plan has not been implemented well so far. The work was done to some extent, but it wasn't further developed. The Nursing Deputy can use these centers as an opportunity." one of the nursing policymakers said in this regard.

Threats

Defects in the country's service delivery system and its not being fully based on the PHC structure as well as the flawed classification and referral system were regard, by participants, as the most important threats.

"The service system in our country is incompletely based on the PHC structure. PHC means the classification of services and the referral system. On the other hand, in case service levels exist and control each other well, we will actually be able to provide appropriate and timely services to the public. Now our nurses have become more specialized, but their only duty is to respond to patients at the time of hospitalization, and they do nothing at the community level before or after hospitalization." one of the health policy makers of the Ministry of Health said on the matter.

DISCUSSION

The present study explained the professional position of community health nursing in the Iranian PHC system from stakeholders' perspective. The extracted areas for the position of community health nursing in the Iranian health system from stakeholders' perspective were classified in 6 main categories including *creating a transparent framework for community health nursing practice*, *enhancing community health nursing education and training for practice in the primary health care system and community settings*, *seeking support*, *strengthening the cooperation and engagement among key stakeholders of the primary health care system*, *changing the policies and structure of the health system* and *focusing on the deficiencies of the health system*. It is worth noting that 4 main categories of this study are consistent with WHO's Community Health Nursing Role Enhancement Model and the main categories *changing the policies and structure of the health system* and *focusing on the deficiencies of the health system* were obtained from the findings of the current study.

The first main category was *creating a transparent framework for community health nursing practice*. In order to create a transparent framework for the practice of community health nursing, a role should be defined for them in the health system and a position, in PHC (20). Therefore, one of the most important infrastructural issues is the development of a position and job description in the organizational chart for community health nurses in comprehensive health centers. Currently, in Iran, the services offered by community health nurses are mainly provided at the third level and in hospitals, because no position exists for community health nurses in comprehensive health centers (4, 28), while in developed countries, the first level of people's contact with the health system is through community health nurses (27). Numerous studies have addressed the need to explain the position of community health nurses in the country's health system and its importance in promoting health and reducing costs (5, 13, 42). Therefore, creating job opportunities for community nurses is among the infrastructure necessary for the provision of nursing services at the community (28).

The second main category was *enhancing community health nursing education and training for practice in the primary health care system and community settings*. One of the important areas of the WHO's Community Health Nursing Role Enhancement Model is the implementation of training programs to empower community health nurses, interdisciplinary training and their continuous professional promotion with the aim of improving the quality of health services provided in health centers (20). Participants referred to the poor community-based education in nursing and argued that that undergraduate curriculum needs to be revised to become community-based. In recent years, due to the increased burden of chronic diseases at the community, nursing education experts around the world aim to make changes in the traditional hospital-based curriculum and approaches, in order to redesign the curriculum with a focus on community-based care (14). The study by Cheraghi et al. also referred to Iranian nursing graduates' insufficient skills and their negative attitudes toward quality care at the community level as a result of being trained through hospital-based services

(43). The educational system of medical universities is not compatible with PHC and curriculums are not tailored to needs, and consequently, university graduates do not possess the necessary skills to face the problems. Therefore, academic education courses should be enhanced and the trainings should be provided in line with the PHC (30, 31). Participants also mentioned the specialization of undergraduate nursing program with a focus on training health nurses tailored to the needs of the health system. In their study, Jarrín et al. stated that the beginning of a community health nursing education program, in the first months of students' arrival, in the form of lectures, introductory textbooks and simulation regarding home-care and community-based care will significantly impact their beliefs and attitude toward community-based nursing, because the traditional curriculum has undermined the value of community activities and home care in students' mind (14).

The third main category covered the development of comprehensive support programs for community health nurses in Iran. In order for support programs to influence the policymakers, a proper understanding of the issues affecting community health nurses and how they relate to people's health at the community is essential. It is necessary to increase public understanding of community health nursing and support for it through the use of mass media (20). Participants referred to the inaction to introduce the community health nursing discipline and its potentials at the academic and community level as an important weakness and considered the development of a communication channel for community health nurses necessary in order to enhance public awareness through the media (30). The study by Heydari et al. also emphasized on preparing the society and increasing the level of public awareness for receiving community-oriented nursing services (28). Poor public perception of their own rights in the health sector is one of the barriers mentioned by the participants. Other studies have also focused on empowering the society to demand health from the government because the members of society do not have the feeling of being a part of the health system and are reluctant to participate in health programs (30). In other words, the lack of discourse between nursing managers and policymakers of the Ministry of Health with the aim of publicizing the capabilities of community health nurses is another weakness been mentioned in this study. The study of Yazdani et al. also showed that there is no mutual discourse between nursing institutions and other institutions of the health system and as a result, the institutions of the health system are not aware of nursing and its master's degrees specialties (44). From participants' perspective, the Nursing Deputy is one of the most effective opportunities to seek support for community health nurses to enter the network system. In response to challenges against the health system, the Ministry of Health established the Nursing Deputy in 2013 (4).

The fourth main category extracted from the study was *strengthening cooperation and engagement among key stakeholders of the primary health care system*. Given the importance of interdisciplinary cooperation, WHO emphasizes the need for the health system to be assessed in each country by its authorities and policymakers in terms of the challenges against interdisciplinary cooperation and engagement (20). From participants' perspective, one of the most important barriers against cooperation and engagement is the sporadic performance of various departments of medical universities. Other studies also refer to problems in interdisciplinary cooperation among different health institutions in the country, ignoring the principle of public engagement and the lack of a clear mechanism for it and failure to utilize the potentials of NGOs and charities that indirectly affect health (29, 30). Intersectoral collaborations in PHC are achieved haphazardly in some areas but they lack a predefined structure and institution (45). However, according to participants, the presence of Nursing Deputy in the Ministry of Health was a good opportunity to strengthen interdisciplinary cooperation with other departments, especially the Health Deputy, for the entry of community health nurses into the network system. In this regard, the study by Yazdani referred to the need for mutual discourse among nursing institutions and other institutions of the health system with the aim of strengthening interdisciplinary cooperation and engagement (44).

The fifth main category of the study was *changing policies and the structure of the health system*. Health care policies in each country are subject to its dominant ideology. In the eyes of health system policymakers, the nursing profession is not taken seriously and is not treated as a valuable profession. The chaos and disorder of the health system has caused people not to fulfill their real roles, and thus the position of nursing and its specialties in the health care system is unclear (44). Participants referred to not giving health priority over treatment in the health system and prioritizing secondary prevention in Iran as one of the most important threats. In line with the results of the present study, other studies have pointed to the treatment-oriented attitude of the managers of the Ministry of Health and the weakened position of health in the health system (30, 44). Participants identified conflicts of interest as another threat to the health system. Other studies suggest that treatment is more attractive than PHC, so physicians and even family physicians adopt a therapeutic approach and are reluctant to offer preventive and care programs (30, 46). From the participants' point of view, another threat is insufficient funding and budgets for recruiting community health nurses in the network system. One of the deficiencies of PHC in developing countries is greater willingness to spend on and invest in specialized and treatment levels (47). In Iran, the PHC system is funded fully by the government and the main weakness of the system is inadequate financing and inconsistency between resources and the necessary service packages. On the other hand, the salary of PHC staff is unfair, not proportionate to the way the services are provided, thus it does not encourage improved performance, quality and efficiency (30, 48).

The last main category extracted in this study was the *deficiencies of health system*. Health care based on the care seekers' needs will mainly lead to health promotion and increased client satisfaction. Thus, health care providers should pay attention to this important matter (49). In interviews with the visitors of comprehensive health centers, they pointed to the routinized care that was provided, and specifically, the middle-aged and elderly visitors were dissatisfied with the consultation and education

received in regard with their underlying diseases. In line with the results of the present study, Heydari et al. also referred the provision of dispersed health services and health experts' not using effective strategies in accordance with the needs of the community (28). Services are provided at a basic level in health centers and people receive supplementary services outside the PHC system, which due to the nature of new needs, they will be difficult to address with this level of services (46).

Another finding mentioned by the participants was the flaw in the PHC-based service delivery system, i.e., failure to follow-up on care seekers at the community level before and after hospitalization as well as patients with chronic diseases or disability under home-care due to the high workload of health care providers and their lack of expertise. In Iran, PHC organizational structure does not have the necessary flexibility for making modifications proportionate to changes and encounters challenges in meeting the new needs of the population, which gradually weakens the system. Therefore, this system needs structural modifications (30, 31, 48). Another threat mentioned by the participants was Behvarzes' lack of response to public health questions due to the increased public knowledge. PHC manpower has not grown in line with the new needs and services and too many duties have been imposed on them. On the other hand, the changes in people's lives and level of knowledge have led to reduced acceptance of Behvarzes and as a result, their relationship with the people has diminished. Thus, recruiting Behvarzes with low level of education is not a good strategy in the current situation. Despite there are university graduates in most of the regions, no mechanism exists to use them (30, 48).

The present study examined stakeholders' perception of the requirements for establishing a community health nursing position in the Iranian primary health care system. Although, community health nursing is not a new concept in Iran and for more than four decades, nursing students have been trained in the field of community health and the Department of Public Health in the past was one of the four nursing departments at level of the Ministry of Health, the transformation and changes in the health system and the physicians' professional dominance have led to the expansion of hospitals. However, the analysis of strengths, weaknesses, opportunities and threats of every main category revealed obvious differences between health system management in Iran and other countries in the promotion of community health nursing. Thus, the Iranian health system have to make management plans and both large- and small-scale policies in accordance with the community health nursing. In this qualitative study, in order to improve generalizability, it was tried to achieve maximum diversity in the selection of participants in the fields of health and nursing policy-making, community health nursing faculty members and those working in health centers from different medical universities across the country as well as visitors. It is suggested that future studies investigate nursing students' perception of the professional position of the community health nursing.

STUDY LIMITATIONS

Covid-19 epidemic made the situation difficult for in-person interviews due to the long distance of some professors, specialists and experts in the field of health across the country. The problem was addressed to a great extent through participants' cooperation and conducting interviews via Skype and What Sapp.

CONCLUSION

Based on the participants' opinions, developing a position for community health nursing in the Iranian health system in line with the provision of community-based health care can be considered as one of the priorities of health system development. It seems necessary to establish community health nursing in the PHC system in order to address the health care needs of the society. Raising public awareness using social networks in regard with the services providable by health nurses can contribute to this goal.

DATA AVAILABILITY STATEMENT

The raw data supporting the results of this article will be available by corresponding author upon request.

ETHICS STATEMENT

This study was approved by the Ethics Committee of Ahvaz Jundishapur University of Medical Sciences (IR.AJUMS.REC.1398.874). The patients /participants provided their written informed consent to participate in this study. Participants were also assured that the data would remain confidential and anonymous. They were also informed that they could be excluded from the study at any time.

AUTHOR CONTRIBUTIONS

AH, SJ, and MR: research conception and design. AH: interview with participants, writing, and drafting of the manuscript. AH, MR, SJ, NE, and SM: analysis and interpretation of data. MR, SJ, NE, and SM: review and editing. All authors contributed to the article and approved the submitted version.

FUNDING

This article is a part of nursing PhD dissertation on nursing from Ahvaz Jundishapur University of Medical Sciences (NO 1398.874) which was financially supported by the Nursing Care Research Center in Chronic Diseases of Ahvaz Jundishapur University of Medical Sciences (NCRCCD-9837).

ACKNOWLEDGMENTS

The authors would like to extend their sincere thanks to the sponsor of the study, participants, managers, and health authorities of the medical universities across the country.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

frontiers in Public Health

ORIGINAL RESEARCH
published: 03 March 2022
doi: 10.3389/fpubh.2021.774864

Knowledge Attitude and Practices of Mitanin's (Community Health Workers) in Chhattisgarh: Malaria Elimination Perspective

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Background: For the success of any program, its implementation plays a crucial role. Community health workers are of immense importance for malaria elimination from India.

Objective: This study was aimed to assess the knowledge gaps and the responsible factors for mitanins' knowledge on various aspects of and problems faced by mitanins during their work.

Methods: Structured interviewer-based questionnaire was used to collect the data, and ordinal regression was applied to analyze the data.

Results: Only 26% of the mitanins were having a good knowledge attitude and practices (KAP) score about malaria. Malaria endemicity of area [odds ratio (OR) = 0.26, 95% CI = 0.13–0.50), $P < 0.001$] and education (OR = 0.35, 95% CI = 0.18–0.69, $P = 0.002$) were the two significant factors affecting the KAP of mitanins.

Conclusion: This study shows that prioritizing education while recruiting the mitanins and training them in the low endemic areas with a focus on malaria, which will help achieve the malaria elimination goal.

Keywords: community health workers (CHWs), malaria, knowledge attitude and practice (KAP), tribals, malaria endemic areas

OPEN ACCESS

Edited by:
Steven W. Howard,
Saint Louis University, United States

Reviewed by:
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Nanloh Jimam,
University of Jos, Nigeria

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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *13 September 2021*
Accepted: *31 December 2021*
Published: *03 March 2022*

Citation:
Ranjha R, Yadav CP, Chourasia MK, Nitika, Dash CK and Kumar J (2022) Knowledge Attitude and Practices of Mitanin's (Community Health Workers) in Chhattisgarh: Malaria Elimination Perspective. Front. Public Health 9:774864. doi: 10.3389/fpubh.2021.774864

INTRODUCTION

India has a target of eliminating malaria by 2030 (1). Malaria control and elimination in rural tribal areas are one of the roadblocks in India's malaria elimination drive (2). The problems in the health sector in tribal areas are compounded by difficult-to-reach areas and poor access to health facilities (3). Several tools utilized for malaria control in India are Long-Lasting Insecticidal Nets (LLINs) and Rapid Diagnostic Tests (RDTs) (2). Although these tools and techniques are of immense importance for malaria elimination, the correct implementation of those tools is the key (4). Due to the huge shortfall of physicians and nurses in the rural and tribal areas, the community health workers (3) (CHW) become the key players for implementing any program.

The malaria elimination program is dependent on LLINs, medicines, and RDTs. These are very good tools for achieving short-term goals. But the pace of malaria elimination in India will depend highly on the skills and knowledge of CWHs. CHWs are an important human resource and contribute significantly to malaria control (5). CHWs are trusted members of the community with a very good understanding of the community (6). They are trusted in the community owing to the same language, ethnicity, and socioeconomic status. The CHWs act as a liaison between healthcare providers and minority/poor communities in rural areas, thus adding value to the healthcare teams. The CHW program in Chhattisgarh is called Mitanin Program. Mitanin in Chhattisgarhi language means friend. The mitanin program was launched in Chhattisgarh in 2002, with the broad objective of providing immediate relief from common health problems and improving health awareness in the rural areas of Chhattisgarh. The village community selects the mitanin in hamlet level meetings and her selection is approved by the village panchayat (Local self govt. body). Currently, there are nearly 60,000 functional mitanins in the state. All the mitanins are supplied with a drug kit which is refilled regularly (7). Mitanins are trained for the community control of malaria and are provided with a guidebook for malaria control and treatment. They act as drug depots to provide the medicines to the patients, carry out the Rapid diagnostic test, and collect blood slides for malaria diagnosis (7).

Chhattisgarh is one of the malaria-endemic states in India. It is inhabited by 2.3% of India's population, but it contributed significantly to malaria morbidity and mortality in India in 2019, 17.8 and 40.3%, respectively (8). The API of the state was 1.97 in 2019. A total of 11 out of 27 districts had an annual parasite index (API) of more than one with the highest API of 44.31 in the Bijapur district (8). More than 30% of the Chhattisgarh population is tribal and lives in forested areas (9). The tribal communities have poor health indicators compared to others. Tribal communities are the most difficult to test and treat (10), thus, increasing the importance of CHWs in these areas. Women and children, being the most vulnerable, are severely affected by different illnesses, and also by lack of awareness on malaria, transportation, discriminatory behavior by healthcare providers, and financial constraints that make CHW of prime importance for delivering health services to them (11). Mitanins being the important and grass root level worker in the health system, their knowledge and skills are important for the effective implementation of health programs. Gaining social recognition, a sense of social responsibility, and self-efficacy motivates the CHWs (12). It is of immense importance from the standpoint of malaria elimination in the country. This study aimed to find out the knowledge gaps and skills of Mitanins in Chhattisgarh and find out the factors that may help in improving the performance of Mitanins for malaria control and elimination in the area.

Abbreviations: ASHA, Accredited Social Health Activist; CHW, community health workers; KAP, knowledge attitude and practices; LLIN, long lasting insecticidal nets; RDT, rapid diagnostic tests; WHO, World Health Organisation.

MATERIALS AND METHODS

Study Area and Study Participants

Chhattisgarh is geographically plain with topographic variations, including plain, foothill, and forested and non-forested areas. This was a cross-sectional study carried out in high malaria endemic as well as low endemic districts of Chhattisgarh. Two subcenter in each district were selected and five villages from the two subcenters were included in the study. An approximate population in each subcenter is 3,000–5,000. There is one mitanin for ∼250 population. The sample size was calculated for the total mitanins in the selected subcenters. It came out to be 200 at a 95% CI and a 5% margin of error. This study was approved by the Institutional Ethics committee ICMR-National Institute of Malaria Research, New Delhi, IEC no- ECR/NIMR/EC/2018/211. Written consent was obtained from the participants in the study. 203 Mitanins were included in this study from April 2018 to December 2019. This study was carried out in 68 villages of 13 districts of Chhattisgarh. The study districts were divided into two groups: low endemic and high endemic (**Figure 1**).

Data Collection and Grading

A structured questionnaire was used to collect information from the mitanins. A pre-tested questionnaire from earlier studies was used for this purpose (13). It was standardized for forested and malaria-endemic tribal regions. Two staff members were trained for the study. The study questionnaire was divided into six sections. The first two sections were regarding the socio-demographics and malaria training-related information. The third and fourth sections were to find out their knowledge about malaria diagnosis and treatment, respectively. The fifth section was focused on their knowledge about malaria prevention with a focus on LLINs. In the last section, documentation by the Mitanins was assessed. The five sections were graded equally, allotting five marks for each section. The total KAP score was calculated out of a maximum of 25 grades. Mitanin knowledge was graded poor, average, and good for <40, 40–70, and $>70\%$ grades.

Data Analysis

All the study data were entered in Epi Info (CDC, Atlanta, Georgia, US). The cleaned data were analyzed using the Statistical Package for the Social Sciences (SPSS) version 20 (IBM Corp, Armonk, NY, USA). All categorical variables were reported as frequency (percentages), and continuous variables were reported as means and SD. Univariate and multivariate ordinal regression analysis was used, and mitanins' KAP score was the dependent variable in the analysis. The mitanins' age, education, work experience, and area endemicity for malaria were used as independent variables. The mitanins' education and experience were dichotomized. Assumptions for ordinal regression were checked for assessing the validity of the regression model. A P-value < 0.05 was considered significant for all the analyses.

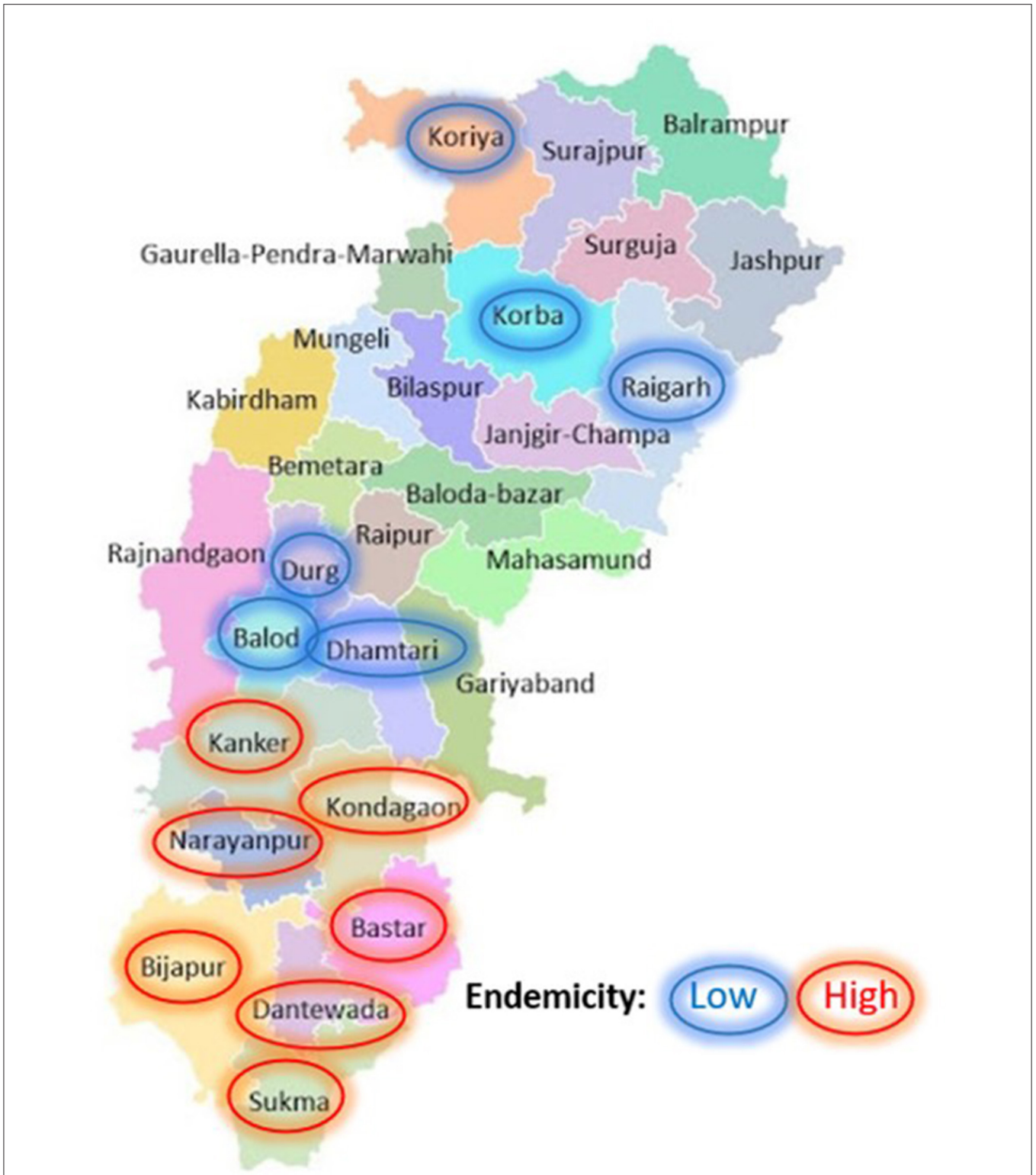

FIGURE 1 | Distribution of selected study districts and their endemicity. The study included mitanin from 13 districts, 6 low endemic (circled blue) and 7 high endemic (circled red) were included in the study.

TABLE 1 | Baseline characteristics and training status of the study population ($n = 203$).

Variable	Category	*n* (%)
Serving area (%)	High endemic	108 (53.2)
Age (in years)		38.54 (9.31)
Education (in years)		5.24 (4.21)
Experience (in years)		11.51 (5.61)
Education (categorized) ($n = 203$)	Less than primary	82 (40.4)
	Primary and above	121 (59.6)
Experience (categorized) ($n = 198$)	Less than 9 years	70 (35.4)
	9–16 years	76 (38.4)
	17 years and above	52 (26.3)
Training status		
Received malaria training	Yes	203 (100)
Regular training on malaria	Yes	185 (92)
Average mitanin per session		26.32 (8.1)
Overcrowded	Yes	38 (18.8)
Training languages	Local	203 (100)
Language problem	Yes	27 (13.4)
Use of different teaching aids	Yes	196 (97)
Content of training	Appropriate	191 (94.1)
Refresher training required	Yes	179 (88.2)

Age, experience, education and average mitanins per session expressed as mean (SD).

RESULTS

Baseline Characteristics and Training Details

A total of 203 Mitanins with a nearly 1:1 ratio based on endemicity were included in the study (**Figure 1**). About half (53%) of the participants were educated until primary level or above. The mean age of the participants was 38.54 (9.31), and the average experience was 11.51 (5.61) years.

All the mitanins were trained, and more than two-thirds of the Mitanin's were willing to take a refresher training on malaria. About 19% of Mitanins felt that the training batches were overcrowded, and about 13% faced language issues. Baseline characteristics and training details are illustrated in **Table 1**.

Mitanins KAP Performance

About 60% of mitanins had an average KAP score**,** about 26% had good KAP scores, and 13.3% had poor KAP scores. For malaria, diagnosis, prevention, and treatment section, the frequency of mitanins with the low scores was high, 76.8, 35, and 24.1%, respectively. Documentation and basic malaria were the sections where the mitanins scores were high (**Figure 2**).

Determinants Affecting Mitanins' Performance

Table 2 shows the results of ordinal logistic regression analysis. Endemicity and education were the two variables affecting the mitanins' KAP score significantly. The proportional odds of higher KAP were 0.26 (95% CI = 0.13–0.50), $P < 0.001$, and 0.35 (95% CI = 0.18–0.69), $P = 0.002$ in the low endemic areas and mitanins with an education level less than primary compared to high endemic areas and mitanins with higher education level respectively. Education was the factor affecting most of the components of mitanins' KAP. The section-wise results of the regression analysis of the mitanin KAP analysis are given in **Supplementary Tables 1–3**.

Problems Faced by Mitanins

About 35% of the mitanins faced problems related to their work; delay in payments and intermittent medicine supply were the two major problems (**Figure 3**). While lack of support from seniors or family members were other common problems reported by the mitanins.

DISCUSSION

Good knowledge of malaria control interventions will ensure better performance of CHWs (14). This study was planned to find out the knowledge of mitanins about malaria control in Chhattisgarh and identify the important factors that may be crucial for improving the knowledge of mitanins.

The mitanins' KAP score was affected by their education [odds ratio (OR) = 0.35, 95% CI = 0.18–0.69, $P = 0.002$] and endemicity [(OR = 0.26, 95% CI = 0.13–0.50), $P<0.001$] of the area being served by the mitanin (**Table 2**).

In low-endemic areas, the odds of high KAP were low. Accredited Social Health Activists (ASHA) in the low-endemic areas had low knowledge about vector breeding, malaria symptoms, and diagnosis compared to counterparts in high endemic areas in northeast India (15). The low knowledge of malaria of mitanins in the low-endemic areas indicates changing focus due to low malaria cases. In the WHO guidelines, surveillance is the core intervention for malaria elimination when the cases go very low (16). The knowledgeable community health workers are of immense importance for making surveillance a core intervention for malaria elimination. Training, monitoring, and assessing mitanins lead to significant improvement in their knowledge from the malaria elimination perspective (17). Separate training sessions on malaria may help fill the knowledge gap of mitanins in the low-endemic areas.

The proportional odds of having a higher KAP were low in the mitanins with low-level education. As per the National Health Mission (NHM), India guidelines for ASHA, a formal level of education up to the 8th class is the criteria for a mitanin's recruitment (18). In our study, only about 19% of the mitanins had an education of 8th class or above. The level of education was reported to be the critical determinant for knowledge of disease and its transmission for individuals involved in integrating community efforts for dengue control (19). Our analysis found that education was the only factor affecting the mitanin's basic knowledge about malaria (OR = 0.39, 95% CI = 0.20–0.80, $P = 0.01$) (**Supplementary Table 1**). The higher education level of CWHs was reported to be associated with good record-keeping, better use of aids, and better counseling (20). Mousoke et al. (21) suggested that the level of education must be given priority while recruiting CHWs. Improving literacy in the tribal areas will positively impact malaria control in the area (22).

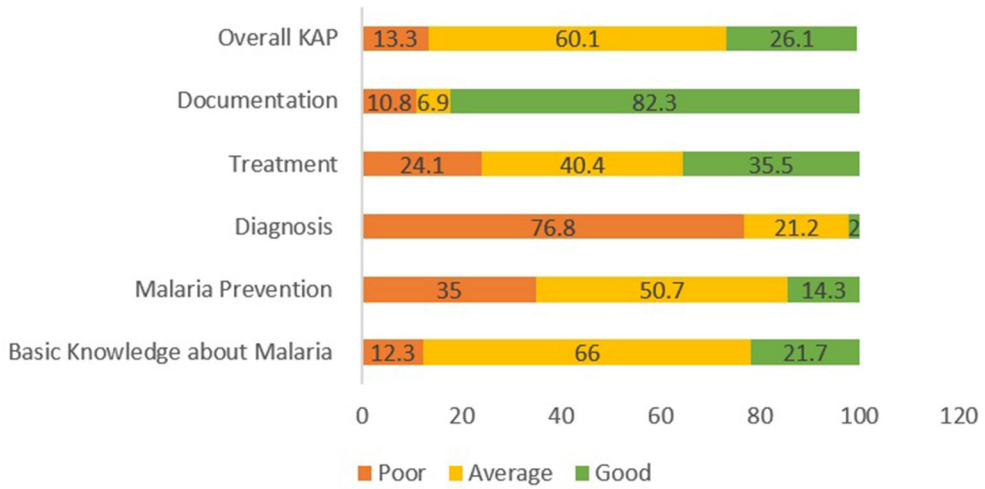

FIGURE 2 | Mitanin KAP Scores. The KAP score of mitanins in different sections and overall scores are highlighted in green (Good), Yellow (Average) and Orage (Poor). Malaria diagnosis, prevention and treatment were the three sections where frequency of low scoring was high.

TABLE 2 | Factors affecting mitanin's performance using ordinal logistic regression ($n = 203$).

Variable	Category	Univariate analysis		Multivariate analysis	
		cOR (95%CI)	*P*-value	aOR (95%CI)	*P*-value
Endemicity	Low	0.33 (0.18–0.59)	**<0.001**	0.26 (0.13–0.50)	**<0.001**
	High	1		1	
Age	In years	0.97 (0.94–1.002)	0.069	0.99 (0.96–1.03)	0.829
Education	Less than 5 years	0.46 (0.26–0.82)	**0.008**	0.35 (0.18–0.69)	**0.002**
	5 years and more	1		1	
Experience	Less than 9 years	2.06 (1.002–4.25)	**0.049**	1.01 (0.41–2.5)	0.97
	9–16 years	2.91 (1.4–6.00)	**0.004**	1.58 (0.72–3.49)	0.258
	17 years and more	1		1	

P-value < 0.05 statistically significant.
Likelihood ratio Chi-square 29.9; <0.001 Pseudo R^2 – 0.16.
Statistically significant values are shown in bold.

Our study supports the earlier observation that mitanins with higher education levels have better KAP. The non-availability of educated individuals may be a factor affecting mitanins recruitment in rural and tribal areas. Also, giving education a priority while recruiting the mitanins may help in the success of the malaria elimination program.

Malaria diagnosis, prevention, and treatment were the three areas where the frequency of mitanins with below-average knowledge was high, 76.8, 35, and 24.1%, respectively. Chowdhury et al. (15) reported a significant difference in the ability to perform by ASHAs in low-endemic and high-endemic areas. 57.24 and 83.16% of ASHAs in the ASAHs in low-endemic and high-endemic areas were able to perform RDT (15). In our study, about 71% of the mitanins correctly told the steps for conducting RDT, but only about 9% could tell the correct time for reading the results but there was no significant difference with the endemicity of the area being served. The low performance in the diagnosis was due to the discrepancy in the RDT protocol and the protocol of the kits provided to the mitanins. Updating mitanins, whenever the kit changed with a new kit having a different protocol, may help in improving the malaria diagnosis by the mitanins.

Mitanins' knowledge about malaria prevention is affected by the endemicity of the area being served. Mitanins of low endemic areas had lower KAP about malaria prevention compared to the mitanins of low endemic areas (OR 0.31, 95% CI 0.17–0.60; $P < 0.001$) (**Supplementary Table 2**). The main malaria prevention tools LLINs and IRS are applied only in the high endemic areas (23). Involvement in the ongoing malaria prevention activities in the area may be the reason for high KAP about

FIGURE 3 | Problems faced by mitanins. Delayed payments and intermittent medicine supply are major problems faced by mitanins.

malaria prevention in the high endemic areas compared to low-endemic areas.

Mitanins' knowledge about malaria treatment is being affected by all the variables considered, including endemicity, education, age, and experience of mitanin (**Supplementary Table 3**). Higher KAP rates about malaria treatment were high in mitanins serving high endemic areas, having higher education and lower age. The low knowledge about malaria treatment in mitanins of low-endemic areas may be due to their dependence on ANM for the same (15). Our study supports the earlier observation that ASHAs in the high endemic areas have significantly better knowledge about malaria treatment compared to low-endemic areas (15). The mitanins with 9–16 years of experience had higher odds of having better KAP about malaria treatment (OR = 22.43, 95% CI = 1.15–5.12; $P = 0.01$). The higher experience of health workers was found to be correlated with better knowledge (24). In our regression analysis, the experience was found to be interacting with education. The frequency of mitanins with primary education and above was higher (∼44%) in the experience level of 9–16 years compared to 17 years and above and <9 years of experience, 23 and 34% respectively. This may be the reason for lower knowledge about malaria treatment in mitanins with experience 17 years and above.

The sustained success of the malaria control and elimination program depends on the data analysis and implementation based on the data (25). Malaria elimination in India will require high-quality real-time data for decision-making, as well as data collection and documentation. The mitanins' performance was good in this section, with 82% of the mitanins having a good score. Mitanins with education level primary and above had good KAP, although it was not significant (OR = 0.43, 95% CI = 0.18–1.04, $P = 0.06$).

Delay in payments and intermittent medicine supply are the two key problems faced by mitanins in the area. Lack of continuous medicine supply may hinder the mitanin from performing their duties effectively. Timely payments are required to keep the mitanins motivated. Smaller, irregular, and delayed payments were problems highlighted in the earlier studies (13, 26). ASHAs/Mitanins were willing to take new responsibilities but expect more incentives. Due to low incentives, they look for other paid jobs along with their healthcare services (27). Getting timely payments may help motivate mitanins for community services.

In conclusion, our study shows that mitanins' KAP performance is affected by the endemicity of the area being served and their education level. The changing focus in low-endemic areas may become a hurdle for bringing malaria cases to zero in those areas. Considering the education of individuals while engaging in the mitanin program may be highly helpful for the long-term goal of malaria elimination. Social and behavioral aspects affecting the utilization of malaria control services in the community can be improved by having a knowledgeable mitanin in the area. Mitanin malaria training should focus on removing the knowledge gap in diagnosing, preventing, and treating malaria. Mitanins in the low-endemic areas needs to be trained more on malaria. Timely payments are required to keep the mitanins motivated. Despite having good diagnostics and effective medicines, there are deaths due to malaria. In 2020, 63 deaths due to malaria were reported in India. Most of the malaria cases and deaths in Chhattisgarh are from rural areas. Mitanins with good knowledge about malaria control may effectively reduce mortality to zero much before malaria elimination from the country.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Institution Ethics Committee, ICMR-National Institute of Malaria Research, New Delhi. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

RR conceptualized the study and wrote the manuscript. RR and CY designed the study. RR, CY, and MC analyzed the data. CY, MC, and N edited the manuscript. CD and JK help in data collection and field coordination during the study. All authors contributed to the article and approved the submitted version.

FUNDING

This study has been funded by National Health Mission, Chhattisgarh.

ACKNOWLEDGMENTS

We are grateful to all Mitanins (CHW) who participated in the study. We would like to acknowledge the State Human Resource Centre, Chhattisgarh, for helping with the study. We also acknowledge the Local Health Departments of different districts of Chhattisgarh for facilitating the study in the corresponding districts. We are grateful to Dr. Khemraj Sonwani, ex-State Programme Officer, NVBDCP, Chhattisgarh for the help in initializing the study.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2021.774864/full#supplementary-material

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ORIGINAL RESEARCH
published: 08 April 2022
doi: 10.3389/fpubh.2022.627169

The Reimbursement Rate of New Rural Cooperative Medical Scheme and Self-Rated Health Among Rural Middle-Aged and Elderly

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Objectives: The ultimate goal of the New Rural Cooperative Medical Scheme (NRCMS) is to improve physical and psychological health and aim to provide equitable, affordable, cost-effective healthcare services for all rural people. One of our major concerns from the perspective of policy outcome is whether middle-aged and elderly can benefit from the insurance to improve self-rated health. The main objectives of this study are to answer the questions that the reimbursement rate of the NRCMS is a possible explanation of why and how rural middle-aged and elderly shift from non-medical service inputs to medical service to produce health based on a family production theory.

Methods: Data were obtained from the China Health and Retirement Longitudinal Study (CHARLS) conducted in 2018, which involved 1,030 rural adults aged 45 years and older, and ordinal logistic regression estimator and two-step regression were used to examine these assumptions. Our approach controlled for the health status of those people at the same administrative level of the hospital.

Results: Our study shows some interesting results. First, the reimbursement rate of NRCMS predicted a higher level of SRH among rural middle-aged and elderly, but that all of the indirect effect of it on SRH could be explained in total by satisfaction of local medical services utilization (ab = 0.0492). Second, the results further showed that the odds ratio of satisfaction from affordable, convenient, high-quality medical services is 2.402 times ($p < 0.01$) greater for those with higher reimbursement levels than for their counterparts with lower reimbursement. Third, the odds ratios of inpatient care visit, outpatient care visit, and physical examination among policyholders of NRCMS are also 1.116, 1.628, and 1.08 times greater, respectively, than their counterparts who are not satisfied with these local medical services.

Conclusions: Our results concluded that generous insurance reimbursement can reduce the price of healthcare and costs of utilization that both had a dramatic effect on SRH among middle-aged and elderly when their demand for medical treatment is incurred. The government should focus on the healthcare cost, utilization, and health benefit calculations of health insurance policy options at the stage of rapid aging in rural China.

Keywords: self-rated health, New Rural Cooperative Medical Scheme (NRCMS), family production theory, reimbursement rate, rural middle-aged and elderly

OPEN ACCESS

Edited by:
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Saint Louis University, United States

Reviewed by:
Linni Gu,
Beijing Normal University, China
Steven W. Howard,
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Specialty section:
This article was submitted to Public Health Policy, a section of the journal Frontiers in Public Health

Received: *07 December 2020*
Accepted: *04 March 2022*
Published: *08 April 2022*

Citation:
Xie X and Hu Y (2022) The Reimbursement Rate of New Rural Cooperative Medical Scheme and Self-Rated Health Among Rural Middle-Aged and Elderly. Front. Public Health 10:627169. doi: 10.3389/fpubh.2022.627169

INTRODUCTION

Rural China is facing an increasingly serious population aging problem. At the end of 2019, the population of elderly people aged 60 years and older increased to about 253.88 million, accounting for 18.1% of China's total population (1). Along with the aging population in rural China, the current health care system and public health insurance benefits for the aged are unable to meet all their needs for better health status. It is well-known that chronic diseases have been the main disease burden of the rural elderly in China. The death toll caused by chronic diseases has accounted for 87% of the total death toll in the country, and its disease burden accounts for about 70% of the total disease burden in China (2). Chronic diseases represent a serious health hazard to rural middle-aged and elderly' physical and psychological health and affect their quality of life, health adjusted life expectancy, and perceived health because of the high prevalence of morbidity, healthcare costs, more treatment cycles, and a high mortality rate. The chronic disease problems among rural older people has rapidly become a major public health challenge in rural China. Improving access to healthcare services for rural middle-aged and elderly is one of the principal objectives of public health insurance. Because rural older adults are often disadvantaged by illness and poverty, enhanced attention on this disadvantaged population is critical to health equity and eliminating poverty (3).

To solve the problem of poor health status and the majority of rural residents who remained uninsured, China launched the New Rural Cooperative Medical Scheme (NRCMS) in 2003 (4). At its inception, the NRCMS was designed to provide public health coverage to most rural residents to improve utilization of health services, prevent rural households from incurring catastrophic health expenditures (CHE), and promote their good health (5). The 2002 State Council Policy Document No. 13 stipulates that the New Rural Cooperative Medical Scheme is a public health insurance system for most rural residents who still lack health insurance (6). Most scholars also refer to the New Rural Cooperative Medical Scheme as 'the NRCMS (7), which contains detailed guidelines for the implementation of rural public health insurance for each participant and manager to follow (8). Moreover, the NRCMS is a premium-based plan financed by a combination of government funds and individual users, with the Chinese government subsidizing 70 to 80% of the insurance premium to overcome poverty and improve access to healthcare services due to rising prices for the insured (9). The reimbursement and generosity of the NRCMS vary by the level of medical facilities: (1) The reimbursement rate for hospitalization expenses is about 90% if the insured visited a village clinic or township hospital or community healthcare center for medical treatment. (2) The reimbursement rate for hospitalization expenses is from 70 to 80% if the insured visited the second-class hospital or county-level hospitals for medical treatment. (3) The reimbursement rate for hospitalization expenses is 50 to 60% if the insured visited tertiary hospitals or downtown hospitals in metropolitan districts for medical treatment (10). Although participation in the NRCMS is voluntary, the NRCMS also requires full household participation or surrender, with either none or all of their folks participating in the social health insurance scheme to reduce adverse selection (11). Whether one has good health or bad health, participation in NRCMS to improve access to healthcare services is one of the main motivations for enrollees.

A key issue in the recent debate over preventing chronic disease in China is whether the extensive coverage of insured individuals by the NRCMS promotes their health (11). Some studies provide evidence that the NRCMS within familiar contexts made a tremendous impact on producing health and improving equity in access to healthcare utilization (12). It is acknowledged that the reimbursement rate is higher for primary health facilities and lowest for city-center hospitals such as tertiary hospitals in order to influence rural middle-aged and elderly to stay close to home and go to their local healthcare centers (13). However, there is the main concern that the rural and other second-tier facilities are viewed by the public as being very low quality, which is not conducive to producing health for middle-aged and elderly in rural China (14). Thus, the generous reimbursement rate of NRCMS aims to modify consumer behavior, encouraging patients to seek medically appropriate levels of care at the appropriate facility, and dissuade them from going to the city-center tertiary hospital simply out of perception that it is always best. There are a few researchers who explored the relationships between social health insurance and self-rated health (SRH) based on specific age groups, and their results showed that social health insurance is strongly associated with health status in different age groups (15). Some studies pointed out that perceived health is strongly influenced by institutional factors such as the healthcare system, healthcare utilization, social health insurance (16). It is universally acknowledged that the ultimate goal of the NRCMS is to improve physical and psychological health for all rural people and is aimed at the provision of equitable, affordable, cost-effective healthcare services at the stage of healthy aging. One of our major concerns from a perspective of implementation effect is whether middle-aged and elderly can benefit from NRCMS in terms of improved access to healthcare services (17).

Thus, many studies have found that NRCMS is closely associated with individual health status through healthcare utilization because it can eliminate the gap in access to health care services and the financial burden of disease in vulnerable groups (18). However, few studies employed healthcare utilization in the analysis of the mediation effect of NRCMS on SRH for middle-aged and elderly. The main aims of this paper were to examine the mediating effects of the satisfaction from the cost, convenience, and quality of local medical services utilization on the relationship between the reimbursement rate for NRCMS and self-rated health among rural middle-aged and elderly. Insights derived from our research may not only contribute directly to

Abbreviations: RRN, reimbursement rate of NRCMS; SRH, Self-rated Health; RRN, reimbursement rate of NRCMS; Medservices, Cost, Convenience and Quality of Local Medical Services; Mastatus, Marital Status; Soactivities, Numbers of Social Activities; Diseases, Number of Chronic Diseases; ADL, Activities of Daily Living; HD, Heart disease; emphysema, Chronic Obstructive Pulmonary Disease; Bca, Bias-Corrected and Accelerated Confidence Interval.

our understanding of the healthcare cost, utilization, and health benefit calculations of health insurance policy options at the stage of rapid aging in China, but also explain how the reimbursement rate of NRCMS impacts SRH by the satisfaction from the cost, convenience and quality of the local healthcare system in rural middle-aged and elderly.

THEORETICAL ARGUMENTS AND FRAMEWORK

Family Production Theory and Healthcare Utilization

In this paper, we aim to examine how the NRCMS's reimbursement rate impacts SRH, applying the Becker family health production model as the theoretical framework (19). Although Mushkin (20), Becker (21), and Fuchs (22) have concluded that an individual's health can be viewed as one form of human capital, no one has researched the influence factor of health production. The traditional economic theory demonstrated that health is the output of human desire, and demand for healthcare is the inevitable outcome of family production (23). People often use a healthy diet, more physical exercise, positive genetic and environmental factors, and cost-effective medical treatment to produce health. Classical demand theory assumed that those hygiene factors above are deemed services and goods purchased in the health market enter consumers' utility function (24). In this approach, the family produces health with the purchase of these market goods such as exercise in the gym, good nutrition from food, more rest time at the expense of working time, etc (25). Some economists have emphasized that family members choose among these market goods based on their relative prices which can include time prices and currency prices. If one good becomes cheaper, we will marginally shift from other more expensive goods to cheaper goods. The "transfer" is marginal-we will use more healthcare service, but less exercise in gym, etc. for instance. Since the most fundamental demand curve is downward-sloping in economics (26), the quantity and quality of the supply of healthcare are usually negatively associated with the market price of healthcare. Bates et al. predicted that the underlying health production function allows for subsidies for medical prices in these markets to alter the optimal amount of health and also stimulate the great demand for cheaper medical service, measured, say, by insurance reimbursement (27). Under this condition, individuals would rather shift from expensive exercise, diet, recreation, leisure or traveling, etc., to utilize cheap healthcare. Social health insurance can reduce healthcare prices compared with other government subsidies; then, lead to a large increase in the quantity of healthcare demand among rural middle-aged and elderly.

Family health production theory states that preventive services are the best input into the production of health but generous health insurance can lower the out-of-pocket price of curative inputs relative to the price of preventive inputs and thereby distort the choice of inputs. We can conclude from those certain conditions above, rising insurance reimbursement rates may simultaneously bring the out-of-pocket medical price down and then increase the quantity of medical care demand. Thus, the insured has much less input of healthy lifestyle into the production of health. The conclusion is consistent with Ehrlich and Becker to a certain extent that the healthy people covered by generous health insurance are often not producing health in the best ways such as going to the gym, early detection of diseases or taking preventive care (28). However, the ex-ante moral hazard effect in healthcare is mostly theoretical because health insurance does not entirely cover the utility loss related to illness (disamenity, pain, disability) because the beneficiary of health insurance still takes the risk of illness and death any time anywhere.

Therefore, the public health insurance among the household may remain attractive because the purchase price of social health insurance is less than the other health inputs, and there is a higher probability of curing illness completely through healthcare utilization. Grossman found that although people inherit an initial stock of health that may be increased by health input, that initial stock depreciates over-time at an increasing rate (29). He asserted that illness or health need, measured by the level of the rate of depreciation that rises with age, definitely be positively associated with medical services utilization (29). Geriatric diseases and biological factors related to aging raise the price of maintaining health and cause the poor and uninsured rural older residents with critical illness to possibly shortened expected lifespan until death is forced to be "chosen". It is a common phenomenon that older people have higher demands for the production of health and desire to purchase health insurance because their stock of health depreciates over-time at an increasing rate. The aging population is a major driver of the rapid annual growth in national health spending and in the demand for healthcare because the overall health among middle-aged and elderly declines with the probability of sickness on the rise. As healthcare consumption in middle-aged and elderly increases, their desire to buy affordable, equal, cost-effective, convenient insurance is grows (30). It has been suggested that health insurance influences the efficiency of the production process of health because it reduces the price of medical care at the point of being sick in the future, so rural older people would like to choose lower premiums but higher reimbursement of insurance. The NRCMS that covers rural residents is their only and best choice. This is really the law of demand (31)- the lower the out-of-pocket medical price, the more people will be willing and able to purchase healthcare services such as inpatient care, outpatient care, physical examinations, etc. Every rural inpatient who is a beneficiary of NRCMS will receive great insurance gains as the government reimburses them for 50% to 90% of total medical costs of hospitalization. Compared to many private medical insurances in China, the New Rural Cooperative Medical Insurance is "a good bargain" for rural older people. If the policyholders of NRCMS fall ill unexpectedly, those people prefer to produce health through insured medical treatment rather than using more expensive inputs, such as exercise, nutrition, etc.

Our research is different from that of others. This study assumed that the reimbursement rate of NRCMS (RRN) impacts SRH among rural middle-aged and elderly through the satisfaction of affordable, convenient, and high-quality

medical services based on family production theory as the underlying framework (**Figure 1**). First, the NRCMS' generous reimbursement can reduce the relative price of healthcare when medical treatment is incurred, so the enrollees would rather choose more affordable medical care to produce health than other inputs when they fall sick. Within the family production framework for examining the effect of a health input on health, gross input in health capital is produced by household production functions whose initial direct inputs also depend on certain "institutional variables" (32). Social health insurance has a positive effect on healthcare cost and utilization. It should be realized in this framework that generous health insurance reduces out-of-pocket medical expenses; so, the insured prefer to shift from nutrition, exercise, recreation, etc. to medical care for producing health. The theory supported the view that generous reimbursement has significantly increased the likelihood of healthcare utilization, such as inpatient care, outpatient care, physical examination, etc. Second, the NRCMS is targeted for improving the accessibility of a medical facility where the insured will get to these medical facilities quickly and conveniently to prepare for producing health in time. Family production theory states that patients will consider spatial factors in examining accessibility to healthcare services (33). Since distance costs are part of the total medical costs among rural middle-aged and elderly, local medical facilities should remove the geographic barrier between the healthcare provider and patients, and those input shifts rely on the efficiency of spatial access (34). The NRCMS aims to provide convenient healthcare and a simple reimbursement approval process, which attracts a lot of middle-aged and elderly to receive medical treatment faster for producing health (35). Last but not least, the anticipated reimbursement rate of NRCMS is lower for the higher-level inpatient medical facilities. This will encourage the local primary medical facilities to provide high-quality medical service as soon as possible to increase profits in the long run. It is common knowledge that local governments and local medical facilities to make every effort to provide comprehensive and high-quality medical services to attract thousands of residents to visit for medical care nearby in order to increase profits or fiscal revenue. So, in that sense, the generous reimbursement rate of NRCMS is positively correlated with the quality of local medical service and then good self-rated health. However, in the context of Chinese medical systems where simpler health needs are handled in the rural local medical facilities and more complex cases are triaged up to the more tertiary hospitals. Thus, in our statistical modeling, we will use health status, administrative level of hospitals and type of medical facility as covariate variables to control the medical appropriateness of going to the city-center tertiary hospital simply out of health need among rural middle-aged and elderly.

Although some studies found that the NRCMS implementation failed to reduce total and out-of-pocket (OOP) medical costs and improve access to healthcare for low-income groups such as rural middle aged and elderly people (36), other studies have found that NRCMS have had a positive impact on reducing healthcare costs and improving access to healthcare among enrollees (37). The effects of NRCMS on healthcare utilization and production of health might be heterogeneous among existing literature since these studies did not demonstrate to the primary beneficiary how to choose among these inputs based on their relative prices. One of the reasons for this is that previous studies would rather use NRCMS' coverage variable than employ insurance reimbursement rates, however, insurance coverage alone cannot comprehensively reflect and measure healthcare costs and utilization. Moreover, these key independent variables are limited measures of insurance claims and do not reflect educating those primary beneficiaries on how to benefit from the NRCMS reimbursement and how much insurance compensation the insured specifically receives from the insurance. Therefore, the independent variable of interest for our study is the reimbursement rate of NRCMS. It is a better substitute for insurance coverage or participation. This study also aimed to examine the assumption that the positive relationship between NRCMS' reimbursement rate and SRH is mediated by the satisfaction from low-cost, convenient, high-quality local medical service utilization among rural middle-aged and elderly.

MATERIALS AND METHODS

Data Sources

This article is a retrospective study based on the China Health and Retirement Longitudinal Study (CHARLS) in 2018 to explore the relationship between the reimbursement rate of NRCMS and SRH in a national sample of rural adults aged 45 years and older. CHARLS is surveyed by the Institute of Social Science Survey of Peking University in China, with the purpose of reflecting a large-scale national, biennial household, and representative survey of China's health, retirement, and population (sampling protocol is publicly available at http://charls.pku.edu.cn/index/en.html). The inclusion criteria of the participants in our analysis were: (1) rural household registration and; (2) aged 45 years and older and;(3) enrolled in NRCMS or Urban and Rural Resident Medical Insurance (URMI) and ; (4) no missing value in all variables. This study removed those samples with missing values because the samples of the CHARLS are large enough. Ultimately, 1,030 respondents were selected, with 837 and 193 in the NRCMS group and URMI group, respectively. All the respondents in the CHARLS provided informed written consent that was in accordance with the Declaration of Helsinki. This paper is a retrospective study based on CHARLS, in which the Ethical Review Committee of Peking University provided ethical approval of the survey (Approval number: IRB00001052-11015).

Variable Definition and Measures

Self-Rated Health

This study adopted the concept of health by WHO as "a state of complete physical, mental and social well-being, and not merely the absence of disease and infirmity" (38). So, the dependent variable of SRH is employed in our article to reflect the outcome of producing health. The dependent variable is self-rated health that was measured by the following question, "Would you say your health is very poor, poor, fair, good or very good?" A five-item scale tapped answers to the question in the CHARLS. This item category scored with 5 points: "1 = very poor", "2 = poor," "3 = fair","4 = good","5 = very good".

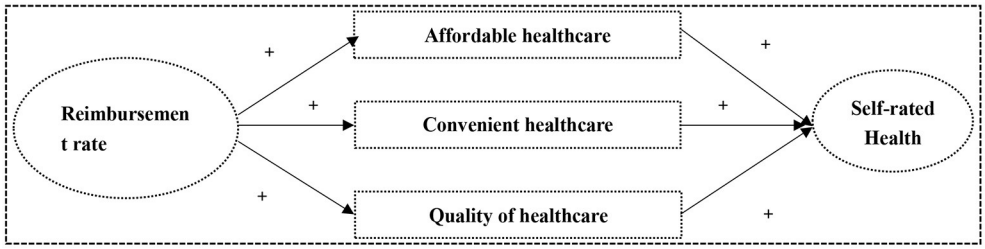

FIGURE 1 | The NRCMS' reimbursement rate impacts the self-rated health among rural middle aged and elderly people through local medical service utilization.

The reimbursement rate of NRCMS (RRN) represents the place where subjects received healthcare services and it is the key independent variable in our analysis. Two items were used to represent the concept of the reimbursement rate in CHARLS. The two items include total medical costs of hospitalization and out-of-pocket costs in CHARLS. First, total medical costs of hospitalization only include fees paid to the hospital, including ward fees but excluding wages paid to a hired nurse, transportation costs, and accommodation costs for yourself or family members. Second, the "out-of-pocket" part was investigated by this question "how much will you pay out of pocket for the total medical costs of hospitalization, after reimbursement from insurance?" Third, we know that total medical costs of hospitalization minus out-of-pocket medical costs equal the reimbursement for medical costs of hospitalization. Thus, RRN = [1- (out-of-pocket medical costs of hospitalization / total medical costs of hospitalization)], which scores on the RRN were coded from 0 to 1.

It should be noted that the mediating variable is adopted in this study to answer the question, "how does RRN impact SRH in rural middle aged and elderly people?". The mediating variable is scored with 5 points is the utilization of local medical services, are you satisfied with the cost, convenience and quality of local medical services when you visited local medical facilities? This item was scored as follows: 1 signified "not at all satisfied", 5 signified "completely satisfied". Three binary variables—whether you have been hospitalized in the past year, whether you visited the medical facility for outpatient care last month, and whether you had a physical examination since the last interview—are employed to reflect healthcare utilization incurred.

Our analysis also controlled for a set of factors that may affect respondents' health conditions and access to healthcare. These included variables for individual social-economic demographics, health status, type and level of health facilities among the respondents, for social-economic demographics, including age, gender, education, marital status, etc. The correct identification of our statistic model that relates an outcome y (SRH) to an independent variable (RRN) relies on the assumption that there is the same or approximate health status among middle aged and elderly people in our sample. Because the scores of activities of daily living (ADL), numbers of disabilities, numbers of chronic conditions, depression, and lifestyle may be proxies for current health status, these sets of covariate variables were controlled in our study. Furthermore, in the context of Chinese medical systems, simpler health needs are handled in the rural local medical facilities and more complex cases are triaged up to the more tertiary hospitals. An obvious concern in our context is critical illness, since patients who received less reimbursement are those who went to the downtown tertiary hospital. Some of these may be more severe cases, and therefore may have reported worse health status. Thus, the second series of binary covariate variables in our study measure whether the subject incurred critical illness, such as stroke, cancer, heart disease, emphysema, etc. In addition, we also controlled the level of this medical facility and the type of this medical facility which patients visited, so, the object of our study is the rural older patients who visited for last medical care at the same type and level of health service facility.

Methodology

Because NRCMS, characteristic of local medical service, healthcare utilization, and SRH are usually discrete variables that are not applicable to stepwise regression referred by Baron and Kenny (39). The two-step logistic regression method that was developed by Zhao et al. (40) who present a nontechnical tutorial in hope of remedying a major defect of the step-by-step procedure method and Sobel z-test proposed by Baron and Kenny (39), to more accurately calculate and robustly test for the mediation effect (40). So, the two-step logistic regression is a good substitute for stepwise regression. In this paper, the statistical process by which RRN affects SRH through local medical service characteristics and healthcare utilization, as shown in **Figure 2**, can be divided into two phases.

Two equations below illustrate that the independent variable X affects the dependent variable Y through the mediating variable M. X is RRN,Y represents SRH, and M represents local medical service utilization and main characteristics (affordable, convenient, and high-quality). The product of regression coefficients a*b represents the indirect impact of RRN on SRH. Two-step logistic regressions estimate the parameters a and b, used to test the indirect effect (mediation effect). In addition, Tofighi et al. (41) recommended that the estimate of indirect effect $u.a \times u.b + \sigma ab$; u.a is the mean of a, u.y is the mean of b (41).

$M = aX + \varepsilon_1$ The first step (Ordinal logistic regression)

$Y = c^{'}X + bM + \varepsilon_2$ The second step (Ordinal logistic regression)

In a nonrecursive three-variable causal model above, Zhao et al. (40) identified two patterns consistent with non-mediation and

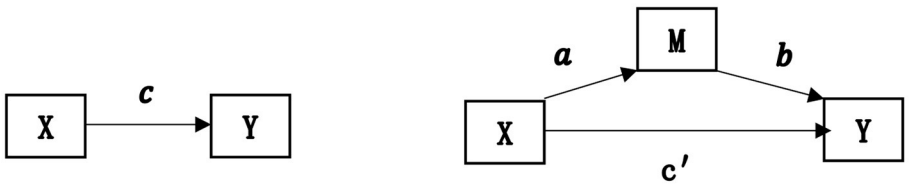

FIGURE 2 | Influence path of the mediating variables. It illustrates that the independent variable X affects the dependent variable Y through the mediating variable M. X is the reimbursement ratio of the New Rural Cooperative Medical Scheme (RRN). Y represents self-rated health. M represents a "local medical service utilization".

three with mediation: (1) No-effect non-mediation: Neither indirect effect nor direct effect exists. (2) Direct-only non-mediation: No indirect effect but only direct effect exists. (3) Indirect-only mediation: No direct effect but only mediated effect exists. (4) Competitive mediation: Direct effect and mediated effect both exist and the two point in opposite directions. (5) Complementary mediation: direct effect and indirect effect both exist, and the two point at the same direction (40).

RESULTS

Table 1 shows a descriptive summary of covariates and major variables categorized by insurance group. We reported the mean and standard deviation of each continuous variable, as well as the observations of each categorical variable. **Table 1** shows the descriptive characteristics of the population of rural middle-aged and elderly. In 2018, the average age of rural middle-aged and elderly was approximately 60.51 ± 9.05 years. **Table 2** is the results of two-way tabulations along with common Pearson's chi-squared that measures of association between different healthcare utilization and satisfaction of local medical services. The results indicated that satisfaction of local medical services are positively correlated with inpatient care, outpatient care and physical examination. Our study used two-step logistic regression to examine the effect of reimbursement of NRCMS on self-rated health in rural middle-aged and elderly.

As shown in **Table 3**, healthy middle aged and elderly people differed significantly from unhealthy middle aged and elderly people in health characteristics and self-rated health. With adjustment by control variables, such as the same health conditions, the same type and level of health service facility visited for medical care, these health characteristics were equally distributed in our ordinal logistic regression between the poor health group and the good health group. So, after controlling for those covariates above, the reimbursement of NRCMS is positively correlated with SRH in rural middle-aged and elderly The odds ratio of RRN is positive but not significant (1.331, $p>0.05$), indicating that the self-rated health reported by some rural middle aged and elderly people with the same health status receiving higher insurance reimbursement may be better than those with less insurance reimbursement. The results held true when our analysis accounted for the beneficiaries' overall health status, type and level of the medical facility they last visited.

The analysis in the theoretical framework stresses that the RRN and SRH usually are not directly connected, but are indirectly related through healthcare utilization. Our results are consistent with most of the studies that lower medical cost was associated with the probability of healthcare utilization and with a higher level of perceived health among the rural older enrollees. As shown in **Table 3**, in a sense, it appears that a high reimbursement rate is positively correlated with local medical service utilization. In rural China, there was significant difference in utilization of local medical services between the low and high reimbursement groups after controlling for their health status, and both level and type of medical facility. Specifically, the results further showed that if a primary beneficiary of NRCMS had higher reimbursement, the odds ratio of the satisfaction from affordable, convenient, and high-quality local medical services utilization was 2.402 times ($p < 0.01$) greater than their counterparts with lower reimbursement. Furthermore, model 4 to model 6 in **Table 3** showed that the odds ratio of inpatient visit, outpatient visit, and physical examination are 1.116, 1.628, and 1.08 times greater, respectively, for rural middle-aged and elderly who are satisfied with local medical service than those unsatisfied with it, controlling for their health status. This finding provides circumstantial evidence that the reimbursement of NRCMS is positively correlated with SRH in rural middle-aged and elderly.

These results supported the family production theory that the generous insurance reimbursement reduces the price of medical care at the point of being ill, so the insured would rather use more medical care than utilize nonmedical services to produce health, when controlling for health status. We discovered that the increase in RRN increased the probability of reporting their good health among rural middle aged and elderly people by controlling health status and health outcomes. In family theory, the quantity of health demanded often declines as healthcare prices rise. This result is consistent with the experience that generous health insurance reduces out-of-pocket expenses for medical care; then, rural middle aged and elderly people covered by generous NRCMS prefer to shift from expensive non-medical services to cheaper medical care when curing diseases is a top priority. In rural China, the probabilities of reporting good health are 1.26 times ($p < 0.01$)greater for middle-aged and elderly who are satisfied with local medical service than those unsatisfied with it, regardless of health status.

Table 4 shows specific indirect effects of the RRN on SRH that is mediated by the satisfaction of affordable, convenient, high-quality local medical services. Our results further found that the RRN predicted a higher level of SRH within these respondents, but that much of the indirect effect of RRN on SRH could

TABLE 1 | Descriptive analysis in rural aged 45 years and older, presented as mean / median, SD, N.

Variables	NRCMS			URRMI		
	N	Mean/Median	SD	*N*	Mean/Median	SD
SRH	837	3.086	0.937	193	2.842	1.021
RRN	837	0.456	0.252	193	0.62	0.286
Medservices	837	3.064	0.997	193	3.375	1.154
Age	837	60.51	9.054	193	59.851	8.370
Male	837	1	0.492	193	1	0.500
Mastatus	837	1	1.315	193	2.097	1.415
Education	837	3.787	1.305	193	2.226	1.190
Sleep	837	1	0.467	193	1	0.488
Smoke	837	0	0.312	193	1	0.246
Drink	837	0	0.438	193	0	0.422
Exercises	837	1	0.285	193	1	0.344
Physical examination	837	1	0.465	193	1	0.499
Soactivities	837	1.704	1.157	193	1.189	0.526
Diseases	837	1.942	1.173	193	1.761	1.043
Disability	837	1	0.417	193	1	0.497
Depression	837	6.367	5.727	193	9.839	6.900
ADL	837	1.791	0.603	193	1.93	0.644
Inpatient care	837	0	0.434	193	1	0.410
Outpatient care	837	1	0.386	193	1	0.375
Hospital type	837	3	0.665	193	4	0.904
Hospital level	837	1	0.793	193	2	0.592
Stroke	837	0	0.268	193	0	0.257
Cancer	837	0	0.145	193	0	0.110
HD	837	0	0.347	193	0	0.284
Emphysema	837	0	0.230	193	0	0.253

RRN, reimbursement rate of NRCMS; SRH, Self-rated Health; Medservices, Cost, Convenience and Quality of Local Medical Services; Mastatus, Marital Status; Soactivities, Numbers of Social Activities; Diseases, Number of Chronic Diseases; ADL, Activities of Daily Living; HD, Heart disease.

be explained in total by satisfaction of cost, convenient, and high-quality local medical services (ab = 0.0492). We generated Bootstrap results for indirect effects from Stata software, and the 95% bias-corrected, accelerated confidence interval (BCa) is 0.0117 to 0.1304. These mediations are significant, because the BCa does not include zero (42). Although the direct effect of the RRN on SRH was about 0.1041, its BCa is from −0.2242 to 0.4255. That BCa obviously includes zero, and is therefore not significant. Therefore, the type of mediation in our study is indirect-only. As expected, our data support the hypothesized mediation story and extended it to family production theory that the RRN affects a distal dependent variable SRH through the satisfaction from local medical service utilization among rural middle-aged and elderly

DISCUSSION

This article is the first to use a nationally representative survey targeting middle aged and elderly people in rural China to explore the relationship between NRCMS' reimbursement rate and SRH based on family production theory. Their mediation effect and health policy implications are discussed here. Our study found that the enrollees would like to use more medical care treatment to produce health when they fall sick, since the NRCMS' generous reimbursement can lower the price of healthcare, improve accessibility and quality of healthcare. It revealed that the reimbursement rate of NRCMS may be viewed as an external economic incentive for rural middle aged and elderly people to produce health by healthcare utilization and health system function. Our results suggested that healthcare costs and utilization had a dramatic and direct effect on better SRH among middle aged and elderly people when their demand for medical treatment is incurred.

Our explanation adopted the family production theory and extends it. Traditional family production theory suggested that rational man marginally shifts from more expensive inputs to cheaper inputs to maintain health. Our findings supported the hypothesis that middle aged and elderly people covered by generous health insurance would rather use more healthcare than non-medical services to produce health when medical treatment is incurred. We demonstrated that the reimbursement rate of NRCMS not only has a significant effect on healthcare utilization but also optimizes local medical facility functions and the degree to which healthcare services for primary beneficiaries

TABLE 2 | Chi-squared test for independence of satisfaction of local medical services and healthcare utilization.

Local medical services	Inpatient visit (*n*)			Outpatient visit (*n*)			Physical examination (*n*)		
	No	Yes	All	No	Yes	All	No	Yes	All
Very dissatisfied	75	19	94	75	19	94	54	39	94
Somewhat dissatisfied	60	15	75	59	16	75	43	32	75
Neutral	383	76	459	379	80	459	259	200	459
Somewhat satisfied	192	43	235	192	43	235	117	118	235
Very satisfied	136	31	167	141	27	167	82	86	167
Chi^2 test	Pearson$chi^2(4) = 9.428$		*P*-value = 0.049	Pearson$chi^2(4) = 12.657$		*P*-value = 0.013	Pearson$chi^2(4) = 47.462$		*P*-value = 0.000

increase the likelihood of desired health outcomes. These findings showed that the higher level of reimbursement rate of NRCMS is positively correlated with low-cost, convenient, and high-quality of local medical services that is expected to lead to a large increase in local healthcare utilization to improve self-rated health among rural middle-aged and elderly So, the main determinant of choice of those health inputs is whether the reimbursement rate of NRCMS raises or lowers their relative prices when demand for medical treatment is incurred among rural middle-aged and elderly. The rapidly increasing geriatric population is not only a major driver in the demand for national health spending and healthcare utilization, but also is susceptible to healthcare costs and their perceived health in rural China. Our study further found that the reimbursement rate of NRCMS not only impacts the employee's choice of health input, but also encourages local medical facilities to provide affordable, accessible, and high-quality healthcare services for those primary beneficiaries. In other words, only generous health insurance has a positive effect on the choice of health input among enrollees or else the utility of going to the gym and running on a treadmill exceeds the utility of going to the hospital and undergoing medical treatment. These findings revealed that a higher reimbursement rate of NRCMS is the external effective institutional economic factor that influences the choices of health input among enrollees. Our research extends family production theory from consumers' utility functions in health economics to social welfare analysis in health policy evaluation. This study has realized that institutional incentives for NRCMS and individuals' rational choices are equally important, and they are closely connected.

These findings provide further scientific evidence for the positive effect of NRCMS on healthcare utilization among middle aged and elderly people in rural China. Our results showed that the odds of middle aged and elderly people who are satisfied with local medical services utilizing inpatient care, outpatient care, and having physical examinations are one to two times greater, respectively, than their counterparts. Our result was consistent with the conclusions of some studies that NRCMS in rural China is the main health financing mechanism to secure access to adequate healthcare service for the insured at an equal, affordable, convenient price to produce health (43). However, our result differs from other studies in that we chose to use reimbursement rates than NRCMS coverage or enrollment indicators. The former option being superior, because it can reflect how those primary beneficiaries benefit from the NRCMS reimbursement and how much insurance compensation the insured specifically receives from the insurance benefit package. There has been much dispute over the question of the policy effect of NRCMS on healthcare costs, utilization and health status based on the differences in data sources, measurement, methodological differences in subject recruitment, and sample bias (44). These differences in age groups may have contributed to the different findings since older adults have a higher propensity for poor health, low incomes, the absence of generous health insurance, under-utilization of high-quality healthcare at city-center tertiary hospitals (45). Furthermore, our analysis also controlled for a set of socio-economic factors that may have impacted access and costs of healthcare utilization among rural middle aged and elderly people, such as health status, age, types of medical facilities, administrative level of medical facilities, etc. Thus, our study might better represent the true association between reimbursement rate of NRCMS and SRH among middle aged and elderly people in rural China than do studies using self-report data on healthy or younger groups without controlling for those demographic and health characteristics.

CONCLUSION

Our study had several limitations. First, since our study draws data for analysis from the cross-sectional 2018 CHARLS, we must be cautious about inferring causal relationships. Secondly, although SRH acts as a health status proxy variable that appears both reproducible and reliable, this variable may also be underestimated or overestimated, because the respondents are often not truly aware of their real physiological and mental conditions. Third, although an individual's perceived health as the dependent variable in our analysis reflects the output from health service utilization, in a sense, the main concern is the degree to which SRH also impacts the person's choice of where they receive their medical care.

Our findings have far-reaching policy implications for promoting equality of NRCMS. First, our results suggest that the

TABLE 3 | The RRN is associated with SRH in rural middle-aged and elderly.

Model	Model 1	Model 2	Model 3	Model 4	Model 5	Model 6
Variables	**SRH**	**Medservices**	**SRH**	**Inpatient care**	**Outpatient care**	**Physical examination**
Medservices			1.260***	1.116***	1.628**	1.080***
			(0.099)	(0.041)	(0.207)	(0.029)
RRN	1.331	2.402***	1.209	1.546**	2.041***	1.337
	(0.438)	(0.788)	(0.405)	(0.327)	(0.423)	(0.282)
Age	1.032***	1.023**	1.029***	1.029***	0.984***	1.060***
	(0.010)	(0.010)	(0.011)	(0.005)	(0.005)	(0.004)
Male	0.874	0.745	0.921	1.145	0.725***	0.796***
	(0.175)	(0.148)	(0.187)	(0.108)	(0.066)	(0.054)
Mastatus (Ref:Married)						
Separation	1.246	1.702	1.242	0.661**	0.893	0.982
	(0.502)	(0.666)	(0.504)	(0.121)	(0.127)	(0.105)
Divorce	0.944	2.846	0.789	1.519	1.226	0.773
	(0.838)	(2.978)	(0.693)	(0.755)	(0.598)	(0.326)
Single	0.598**	0.828	0.614**	1.050	1.112	0.926
	(0.135)	(0.184)	(0.139)	(0.109)	(0.114)	(0.072)
Education (Ref: Illiterate)						
Primary school	0.706	1.024	0.693	1.038	0.939	1.211**
	(0.186)	(0.269)	(0.184)	(0.126)	(0.110)	(0.107)
Middle school	0.603**	0.717	0.606*	1.270*	1.072	1.498***
	(0.155)	(0.183)	(0.157)	(0.156)	(0.127)	(0.135)
High school	0.432***	0.576*	0.459***	1.296*	1.231*	1.404***
	(0.125)	(0.169)	(0.134)	(0.178)	(0.155)	(0.135)
Associate degree	0.601	0.486*	0.622	1.266	1.364*	2.309***
	(0.243)	(0.191)	(0.254)	(0.247)	(0.235)	(0.299)
Bachelor's degree and Above	1.752	0.407	1.923	2.480	3.508**	4.221***
	(2.232)	(0.422)	(2.458)	(1.623)	(1.791)	(2.165)
Soactivities	0.956	0.956	0.965	1.002	1.178***	1.221***
	(0.115)	(0.113)	(0.117)	(0.060)	(0.057)	(0.047)
Sleep	1.037	1.233	1.033	0.984	1.126	1.287***
	(0.176)	(0.209)	(0.176)	(0.082)	(0.088)	(0.075)
Drink	0.902	1.115	0.899	1.397***	1.063	0.969
	(0.202)	(0.248)	(0.202)	(0.150)	(0.107)	(0.069)
Smoke	0.850**	0.567**	1.165	1.072	0.846	0.166***
	(0.054)	(0.148)	(0.209)	(0.082)	(0.129)	(0.065)
Exercises	1.574	0.982	1.629*	1.007	1.243	1.300**
	(0.447)	(0.277)	(0.465)	(0.141)	(0.184)	(0.139)
Diseases	0.768	0.936	0.765	1.846***	1.546***	1.440***
	(0.148)	(0.184)	(0.148)	(0.172)	(0.132)	(0.094)
Disability	0.740*	1.020	0.745*	1.102	0.979	1.086
	(0.120)	(0.178)	(0.121)	(0.092)	(0.083)	(0.075)
Depression	0.904***	0.964***	0.909***	1.016**	1.010	0.984***
	(0.012)	(0.012)	(0.012)	(0.007)	(0.006)	(0.005)
ADL	0.538***	0.903	0.542***	1.223***	1.288***	0.921
	(0.078)	(0.128)	(0.079)	(0.091)	(0.090)	(0.051)
Stroke	0.552*	0.675	0.578*	1.539***	0.979	1.290*
	(0.168)	(0.203)	(0.176)	(0.250)	(0.176)	(0.196)
Cancer	0.596	0.772	0.602	2.833***	1.199	1.168
	(0.234)	(0.297)	(0.237)	(0.813)	(0.377)	(0.327)

(Continued)

TABLE 3 | Continued

Model	Model 1	Model 2	Model 3	Model 4	Model 5	Model 6
Variables	**SRH**	**Medservices**	**SRH**	**Inpatient care**	**Outpatient care**	**Physical examination**
HD	0.649	0.687	0.639	0.928	0.797	1.445***
	(0.196)	(0.204)	(0.194)	(0.146)	(0.127)	(0.187)
Emphysema	0.897	1.072	0.874	1.779***	1.638***	1.189
	(0.230)	(0.279)	(0.226)	(0.244)	(0.224)	(0.149)
Hospital level (Ref: County/District hospital)						
Regional/City hospital	0.815	0.791	0.854	1.449***	1.247***	0.998
	(0.179)	(0.173)	(0.190)	(0.130)	(0.107)	(0.063)
Provincial/Affiliated to a ministry hospital	1.234	0.860	1.331	2.127***	0.944	0.571***
	(0.378)	(0.271)	(0.409)	(0.151)	(0.118)	(0.074)
Military hospital	0.822	1.401*	0.804	1.184*	0.922	0.769**
	(0.153)	(0.266)	(0.150)	(0.104)	(0.076)	(0.088)
Hospital type (Ref: Village clinic/Private clinic)						
Health care post	0.942	0.982	1.080**	1.006	1.058	0.716
	(0.100)	(0.043)	(0.036)	(0.031)	(0.152)	(0.165)
Township hospital	0.825	1.299	1.543***	0.681	1.051	0.998
	(0.186)	(0.273)	(0.225)	(0.853)	(0.045)	(0.081)
Community healthcare center	0.943	0.131***	1.745	0.653**	1.352**	0.876
	(0.583)	(0.060)	(0.718)	(0.137)	(0.199)	(0.188)
Specialized/Chinese medicine hospital	0.660***	0.902	0.979	1.425***	0.594***	1.429*
	(0.080)	(0.114)	(0.085)	(0.187)	(0.090)	(0.304)
General hospital	0.731*	0.797**	1.100	1.254**	1.051	0.998
	(0.126)	(0.071)	(0.066)	(0.114)	(0.045)	(0.081)
Constant				0.011***	0.212***	0.008***
				(0.005)	(0.090)	(0.003)
Observations	1,030	1,030	1,030	1,030	1,030	1,030
Pseudo R-squared	0.102	0.0440	0.109	0.109	0.0613	0.0642

*RRN, reimbursement rate of NRCMS; SRH, Self-rated Health; Medservices, Cost, Convenience and Quality of Local Medical Services; Mastatus, Marital Status; Soactivities, Numbers of Social Activities; Diseases, Number of Chronic Diseases; ADL, Activities of Daily Living; HD, Heart Diseases. Robust standard errors in parentheses.*** $p < 0.01$, ** $p < 0.05$, * $p < 0.1$.*

TABLE 4 | Build BCa and test mediation hypotheses using the bootstrap method.

RRN > LMS > SRH	Coef	Bias	SE	95% BCa	Significant
Indirect effect	0.0492	0.0011	0.0285	0.011727 0.130436	Yes
RRN > SRH					
Direct effect	0.1041	0.0027	0.1605	−0.2242029 0.4254853	NO

BCa, Bias-corrected and Accelerated Bias corrected Bootstrap.

next public health reform in rural China should replace Fee-for-Service (FFS) with Diagnosis Related Group Systems (DRGs)-based reimbursement to achieve dual goals: cost containment and maximization of health output. Second, the reimbursement rate of hospitalization expenses should be increased in middle aged and elderly people at a disadvantage of health and economic status after the NRCMS merged with URMI came into effect. Third, the government should devote its attention to providing more affordable, cost-effective, convenient, and equitable health insurance for middle aged and elderly people who would like to use more medical services to produce health when their medical treatment is incurred in rural China. Last but not least, the government not only needs to focus on the NRCMS' design, implementation and reform, but also emphasize the fact that the health care costs and utilization are the vital nexus between the NRCMS and SRH that is a very important policy mechanism to produce health in a public health system. How to reduce the burden of disease and produce better health for rural middle aged and elderly people by bringing the NRCMS benefits of the package is one of the key priorities of rural China's public health insurance reform at the stage of rapid aging of the population.

DATA AVAILABILITY STATEMENT

The dataset supporting the conclusions of this article is available in the http://charls.pku.edu.cn/pages/data/2018-charls-wave4/en.html.

ETHICS STATEMENT

All the respondents in the CHARLS provided informed written consent that was in accordance with the Declaration of Helsinki. This paper is a retrospective study based on CHARLS, in which the Ethical Review Committee of Peking University provided ethical approval of the survey (Approval number:IRB00001052-11015).

AUTHOR CONTRIBUTIONS

XX conceived, designed, and conducted the original research, who acquired and analyzed the data. Drafting the work or revised it critically about nearly 70% was a substantial contribution to XX. YH contributed to the methods, collection and interpretation of data, and proofreading of final paper together. Both authors contributed to the article and approved the submitted version.

FUNDING

This study was funded by General Projects of the National Social Science Fund of China from 2018 to 2021(Approval Number: 18BZZ101).

ACKNOWLEDGMENTS

We acknowledged that the Institute of Social Science Survey of Peking University in China give us official permission to download and use the data of CHARLS.

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Conflict of Interest: YH was employed by West China Hospital, Sichuan University.

The remaining author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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