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Hospital management and healthcare policy: Financing, resourcing and accessibility

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Hospital management and healthcare policy: Financing, resourcing and accessibility

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Editorial: Hospital management and healthcare policy: financing, resourcing and accessibility

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Editorial on the Research Topic
Hospital management and healthcare policy: financing, resourcing and accessibility

As the editors of "Frontiers in Public Health," Research Topic "*Hospital Management and Healthcare Policy: Financing, Resourcing and Accessibility*," we are pleased to introduce this Research Topic, which highlights pivotal research across multiple critical categories: Healthcare policy, Hospital Management, Healthcare Financing, Healthcare Information, and Health Insurance. Each of these domains represents a cornerstone in the architecture of effective public health systems worldwide. This collection of studies provides invaluable insights into the challenges and innovations shaping the future of global health.

Healthcare policy

Healthcare policy remains at the forefront of addressing disparities and optimizing care. One compelling study, "*Association of geographical disparities and segregation in regional treatment facilities for black patients with aneurysmal subarachnoid hemorrhage in the United States*," (Kabangu et al.) underscores the urgent need to address regional inequalities in healthcare access. Additionally, "*The socioeconomic burden of spinal muscular atrophy in Saudi Arabia: A cross-sectional pilot study*" (Alotaibi et al.) illuminates the profound economic impact of rare diseases on families and health systems.

Cost-effectiveness analyses, such as those conducted for tobacco cessation interventions in Thailand and atezolizumab treatment for non-small-cell lung cancer in the UK, offer critical evaluations of resource allocation in health interventions. The study on corruption types during the COVID-19 pandemic across Western and Central-Eastern health systems provides a stark reminder of the systemic challenges that can undermine public health efforts.

Moreover, the research on inpatient respiratory conditions before and during the COVID-19 pandemic reveals significant changes in healthcare demands and patient profiles. A systematic review and meta-analysis on financial toxicity among cancer patients

highlights the economic burdens faced by individuals across different income countries, emphasizing the need for equitable healthcare financing.

Hospital management

Hospital management plays a vital role in ensuring the efficiency and effectiveness of healthcare delivery. The comparative study of human resource management in the top five global hospitals offers valuable lessons in achieving operational excellence. In China, a DEMATEL-Based network analysis model identifies key factors affecting the mental health and performance of hospital administrators working from home, reflecting the new challenges posed by remote work environments.

A machine learning-based cross-sectional study on the healthcare costs of cardiovascular disease in China and a micro-costing analysis of suspected lower respiratory tract infection care in a French emergency department provide deep dives into the financial intricacies of healthcare services. Qualitative insights from Chinese hospital leaders on Joint Commission International (JCI) accreditation and a game theory-based approach to optimizing internal control in public hospital supply chains further contribute to the discourse on hospital management.

Healthcare financing

In healthcare financing, innovative approaches to risk governance, such as those addressing fraudulent reimbursement of patient consultation fees in China, are critical in maintaining the integrity of healthcare systems. The consideration of patient perspectives in economic evaluations of health interventions in Canada represents a progressive step toward patient-centered care. Additionally, the cross-sectional comparison of healthcare delivery and reimbursement between segregated and non-segregated communities in Hungary sheds light on the implications for addressing social and economic disparities of healthcare access.

Healthcare information

The effective use of healthcare information systems is essential for modern healthcare. Studies on incentive mechanisms for sharing and using electronic health records (EHR) in medical consortiums and the application of telemedicine systems for older adult postoperative patients in community settings highlight the transformative potential of digital health innovations. These studies from China emphasize the importance of performance evaluation and feasibility in implementing new health technologies.

Health insurance

Health insurance is a fundamental pillar of universal health coverage. The appraisal of universal health insurance and maternal health services utilization before and after the implementation of Jaminan Kesehatan Nasional (JKN) in Indonesia provides critical insights into the impact of health insurance policies on healthcare access and outcomes. This study exemplifies the ongoing efforts to achieve health equity through comprehensive insurance schemes.

In conclusion, this Research Topic brings together a diverse array of studies that collectively advance our understanding of healthcare policy, hospital management, healthcare financing, healthcare information, and health insurance. Each article offers its unique contributions to the ongoing discourse on how to improve public health systems globally. As we continue to face new challenges and opportunities in public health, the research presented here serves as a vital resource for policymakers, practitioners, and scholars dedicated to fostering healthier communities worldwide.

Author contributions

B-LW: Conceptualization, Validation, Writing – original draft, Writing – review & editing. JS: Conceptualization, Writing – review & editing. HH: Conceptualization, Writing – review & editing. JH: Conceptualization, Writing – review & editing. TW: Conceptualization, Supervision, Validation, Writing – review & editing.

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Cost-effectiveness analysis of atezolizumab in patients with non-small-cell lung cancer ineligible for treatment with a platinum-containing regimen: a United Kingdom health care perspective

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Background: Cost-effectiveness of atezolizumab, as a treatment for advanced non-small-cell lung cancer (NSCLC) patients who cannot receive a platinum-containing regimen,was still unknown. Our objective was to evaluate the cost-effectiveness of atezolizumab vs. chemotherapy in this indication from the perspective of UK healthcare system.

Methods: From the global, randomised, open-label, phase III IPSOS trial, clinical inputs and patient characteristics were obtained. A partitioned survival model with three health states was built: Progression-free survival, progressed disease and death. A lifetime time horizon was applied, with an annual discount rate of 3.5%. Additionally, the willingness-to-pay threshold of £50,000/QALY was utilized. Primary outcomes were quality-adjusted life-year (QALY), costs, and incremental cost-effectiveness ratio (ICER). Sensitivity, scenario, and subgroup analyses were used to assess the reliability of base-case results. Price simulations were carried out in order to provide information for the pricing strategy at specific willingness-to-pay threshold.

Results: In the base-case analysis, atezolizumab resulted in a gain of 0.28 QALYs and an ICER of £94,873/QALY compared to chemotherapy, demonstrating no cost-effectiveness. Price simulation results revealed that atezolizumab would be preferred at a price lower than £2,215 (a reduction of 41.8%) at the willingness-to-pay threshold of £50,000. Sensitivity, scenario and subgroup analyses revealed these conclusions were generally robust, the model was most sensitive to the price of atezolizumab and subsequent medication. Furthermore, atezolizumab was found to be more cost-effective for patients displaying a positive PD-L1 expression, with an ICER of £72,098/QALY as compared to chemotherapy.

Conclusion: Atezolizumab is not cost-effective for patients with advanced NSCLC ineligible for platinum-containing regimen, potential price reduction is necessary.

KEYWORDS

atezolizumab, non-small-cell lung cancer, UK, platinum-ineligible, cost-effectiveness, price simulation

1. Introduction

Globally, lung cancer is the second most frequently diagnosed cancer and is responsible for the most of cancer-related deaths. In the UK, lung cancer accounts for 13% of newly diagnosed cancer cases and is associated with 21% of cancer-related deaths (1, 2). With the process of aging, the prevalence of lung cancer has been on the rise. Non-small-cell lung cancer (NSCLC) accounts for the largest proportion among all types of lung cancer, with a staggering 88% prevalence. Additionally, over half of the patients are already in advanced stages at the time of diagnosis (3). Consequently, there is a substantial burden associated with lung cancer. In 2010, the overall expenses of lung cancer over a span of 5 years amounted to around £267 million in the UK. When considering value-based oncology, it becomes crucial to assess the relative cost-effectiveness of various treatment options (4).

For NSCLC patients with negative driver genes, the current standard treatment is platinum-based doublet chemotherapy, combined with immunotherapy and/or anti-angiogenic therapy (5). However, in the real clinical setting, a significant portion of patients cannot tolerate platinum-based chemotherapy. Initially, the majority of NSCLC patients in the real world are diagnosed with an Eastern Cooperative Oncology Group (ECOG) performance status (PS) score of ≥2. In the UK, it is estimated that 53% of lung cancer patients have an ECOG PS score of ≥2 (6). In most of clinical studies focused on immunotherapy, ECOG PS-high-score and older adult patients were excluded (7, 8). Regardless of the type of treatment received, patients with an ECOG PS score ≥ 2 had a worse prognosis compared to patients with a PS score of 0–1 (9–11). Secondly, statistics showed that the average age of onset for NSCLC patients is >70 years old (1). Overall, these patients usually have some comorbidities or contraindications that make them unsuitable for platinum-based chemotherapy. For NSCLC patients who cannot tolerate platinum-based chemotherapy, recommended treatments included combination therapy, monotherapy, or palliative care (12). With such treatments, the median survival time for patients was only 9.2–9.5 months (13, 14). Therefore, it is necessary to investigate strategies that offer enhanced effectiveness and safety for individuals within these patients.

The IPSOS trial is the first and only global phase III randomized controlled validation study conducted in a population not suitable for receiving platinum-based doublet chemotherapy (14). In 23 countries across Europe, Asia, and America, the research was carried out in 91 regions. Patients who met the criteria were randomly divided into two groups, with a ratio of 2:1. One group received atezolizumab ($n = 302$), while the other group received chemotherapy ($n = 151$). The objective of this study was to assess the efficacy and safety of atezolizumab as the initial treatment among these patients (more details are provided in Supplementary Table S1). The results showed that atezolizumab significantly reduced the death risk by 22% and also decreased the risk of disease progression by 13%, which suggested that atezolizumab was a potential first-line choice for advanced NSCLC patients who cannot undergo platinum-based chemotherapy.

Atezolizumab has been approved in the UK for the treatment of five indications of NSCLC (15). (1) Adjuvant treatment for patients with stage II to IIIA NSCLC who have a high PD-L1 expression level. (2) It is recommended to be added to bevacizumab, paclitaxel, and carboplatin or nab-paclitaxel and carboplatin, as the first-line treatment for patients with advanced non-squamous NSCLC. (3) The initial therapy for adult patients with metastatic NSCLC having PD-L1 expression of 10% or more is recommended. (4) Second-line treatment for patients diagnosed with locally advanced or metastatic NSCLC, who have undergone chemotherapy previously. The disease burden for the patients of interest is significant (6), considering the IPSOS trial results were published in July 2023 in the Lancet, it is expected that the indication of atelizumab for platinum-based chemotherapy intolerant population would be expedited for approval in the UK (14). In order to offer new, effective, and safe treatment options for advanced NSCLC patients who are ineligible for treatment with a platinum-containing regimen, as well as provide a more economical solution to alleviate their disease-related financial burden and ensure the optimal allocation of limited health resources, clinicians and decision-makers need information on cost-effectiveness to make informed healthcare decisions. Therefore, we aimed to inform decision-makers about the cost-effectiveness of atezolizumab in the UK healthcare system. The analysis was conducted from the UK healthcare system perspective to provide evidence for health technology assessment submissions and establish drug pricing strategies.

2. Materials and methods

2.1. Model overview

The purpose of this cost-effectiveness analysis was to compare the clinical and economic outcomes of atezolizumab with chemotherapy in patients with advanced NSCLC who cannot receive a platinum-containing regimen.

A partitioned survival model was created, comprising three health states: progression-free survival (PFS), progressed disease (PD), and death (Supplementary Figure S1). A 10-year lifetime horizon was applied. The percentage of patients who were alive or free from progression at each cycle (cycle length: 21 days) were estimated using the areas under the OS or PFS curves. By calculating the difference between the OS and PFS curves, the PD rate was determined. In the PFS state, patients were administered treatment using either atezolizumab or single-agent chemotherapy (vinorelbine or gemcitabine). Once the disease progressed or drug-discontinuation, patients would receive subsequent treatments consistent with the IPSOS trial (14). For patients receiving no subsequent anti-cancer

treatments, best supportive care (BSC) was performed, more details are available in the Supplementary Table S2.

According to the National Institute for Health and Care Excellence (NICE) reference case, the model was developed from the aspect of the UK National Health Service (NHS) and personal social services (16). Both costs and utilities were discounted annually at a rate of 3.5% (17). The threshold for willingness-to-pay was established at £50,000 for each quality-adjusted life-year (QALY), taking into consideration the population of interest receiving "end-of-life" treatment, as recommended by the NICE (18). Additional analyses were conducted using thresholds of £30,000 per QALY and £90,000 per QALY. The reported outcomes included costs, life-years, QALY, and incremental cost-effectiveness ratio (ICER).

2.2. Population and health state transitions

Clinical inputs and patient characteristics were all extracted from the IPSOS trial (14). The population of interest were individuals with stage IIIB or stage IV NSCLC, who were ineligible for platinum-based therapy, had an ECOG PS of 2–3, mean age of 75 years, and possessed wild-type EGFR or ALK gene mutations. Besides, males comprised 73% of the population. In addition, according to a NICE technology appraisal (17), the parameters assumed for the population in this model were a mean weight of 74.1 kg, an average height of 170 cm, and a body surface area of 1.85 m^2.

The OS and PFS probabilities were derived from the Kaplan–Meier curves documented in the IPSOS trial (14, 19). The required data was extracted using the GetData Graph Digitizer software. Guyot's method was utilized to reconstruct the estimates of individual patient data (IPD) (20). The accuracy of the IPD reconstruction was assessed using the root mean square error (RMSE). The RMSEs of reconstructed IPDs were ranging from 0.002 to 0.006, which indicated a high accuracy. Subsequently, the reconstructed IPD was utilized to fit various parametric functions, including exponential, gamma, Gompertz, Weibull, generalized gamma, log-normal, log-logistic, fractional polynomial (FP), restricted cubic spline (RCS), and Royston-Parmar spline (RP) models. The goodness-of-fit criteria for model selection were evaluated based on the Akaike information criterion (AIC) and visual inspection (21).

Furthermore, age-based adjustments were made to the mortality rate to ensure it would not fall below that of the general population in the UK (22).

We opted for the Gompertz model for OS and RP-hazard models for PFS of atezolizumab. As for the PFS and OS of chemotherapy, we selected the RP-odds and Log-normal models. Detailed parameters are presented in Supplementary Table S3. Further information of goodness-of-ft results can be found in Supplementary Table S4 and Supplementary Figure S2.

2.3. Adverse events

Adverse events (AEs) rates were extracted from the IPSOS trial (14), considering only AEs of grade 3 or higher with an incidence exceeding 1% in either group. These events encompassed dyspnoea, anemia, neutropenia, leukopenia, nausea, vomiting, rash, decreased white blood cell count, and decreased neutrophil count. For additional details regarding incidences, durations and costs, refer to Supplementary Tables S5, S7.

2.4. Treatment duration

As per the clinical trial design, we assumed that treatment would continue until disease progression or unacceptable toxicity occurred. Median treatment duration for patients receiving atezolizumab and chemotherapy were 6 and 4 cycles, respectively. Observations revealed discontinuation rates of 13% for atezolizumab and 14% for chemotherapy (14). Since specific discontinuation times were not available for each patient, we employed the DEALE method to estimate the cyclical rate (23). The cyclical discontinuation rates for atezolizumab and chemotherapy were 2.3 and 3.7%, respectively.

2.5. Health state utilities

A disutility approach was used, which took into account the decrease in utility as age and gender increase, based on the population norms of the UK EQ-5D-3L (24). The decrement can be summarized as follows:

$$\text{Utility} = 0.951 + 0.0212 \times \text{Male} - 0.000259 \times \text{Age} - 0.000032 \times \text{Age}^2$$

The reported utility values in each study were used to calculate the disutility associated with each health state by subtracting it from the general population utility. Then, these disutility values were deducted from the population norms. The base-case analysis utilized the disutilities reported from IMpover150 (25), a study that examined the treatment of metastatic non-squamous NSCLC with atezolizumab. Using EQ-5D questionnaire, The utility values for PFS and PD when undergoing treatment were calculated as 0.71 and 0.69, respectively. The disutilities of PFS and PD when not receiving treatment were obtained from van den Hout's research (26). Likewise, it was observed that the patients in van den Hout's study adequately represented the population of interest. Disutilities of AEs were included, and values were all from NICE committee papers, the duration-adjusted negative effects caused by AEs were assumed to occur during the initial cycle (27–29). More details can be found in Supplementary Tables S6, S7.

2.6. Treatment unit costs and health state costs

Only direct medical costs were considered in our study, encompassed the costs of acquiring active-treatment drugs and follow-up items, along with the expenses of AE management, BSC, and end-of-life care. The prices of generic drugs were obtained from the electronic market information tool (eMIT) for the year 2022 (30). Prices of medications were obtained from the listed price outlined in the 2022 British National Formulary (BNF) (31). We assumed complete vial sharing for all weight-based medications, as a conservative estimate (4). In this study, we utilized the NHS 2021–2022 reference cost (code SB12Z) (32) to calculate administration costs (£287/cycle) for all intravenous drugs (See more

in Supplementary Tables S8, S9). Due to the lack of reported by the IPSOS trial regarding the usages of vinorelbine and gemcitabine, we assumed that in our base-case analysis, 50% of the patients received intravenous vinorelbine, while the remaining patients received gemcitabine. Besides, dosing intensity was assumed to be 100% for both groups due to lack of report. Healthcare resource utilized during the follow-up period included CT chest scan, chest radiography, electrocardiogram, outpatient visit, community nurse, clinical nurse specialist, general practitioner surgery, general practitioner home visit, and therapist visit. Prices of follow-up items were obtained from 2021–2022 NHS reference costs and the 2022 Personal Social Services Research Unit (PSSRU) costs (33). Follow-up care costs were £326 and £506 per cycle for PFS and PD in this model, respectively, more information is available in Supplementary Table S10. AE management costs were obtained from NHS or NICE committee papers targeting on advanced NSCLC (Supplementary Table S5) (17, 34). BSC was consisted by radiotherapy, morphine, bisphosphonate, steroids, nonsteroidal anti-inflammatory drugs, denosumab and dietitian, doses and prices of above items were taken from NHS, BNF or eMIT. In our base-case analysis, cost for BSC was £379 per cycle. The costs of end-of-life care was considered a one-time expense. Based on a NICE committee paper (17), the average cost per episode of end-of-life care in our model was £4,773, details for costs of BSC and end-of-life care are available in Supplementary Table S11. The prices of all mentioned items were adjusted to 2022 using the PSSRU annual inflation hospital and community health services index.

2.7. Sensitivity analyses

We conducted deterministic sensitivity analysis (DSA) to examine the impact of crucial parameters on the ICER, and the findings were presented as tornado diagrams. All parameters were modified either within the designated 95% confidence intervals (CI) or by ranging the base-case values (±20%). Detailed sources of uncertainty are provided in Supplementary Table 12.

A Monte Carlo simulation was performed with 10,000 iterations to conduct probabilistic sensitivity analysis (PSA) on the base-case. Additionally, we conducted 1,000 iterations for the PSA of scenario and subgroup analyses. For cost, we opted for the gamma distribution, while for probability, proportion, and utility, we chose the beta distribution. The scatter plots were utilized to visually present the outcomes of the base-case PSA. Afterwards, probability of being cost-effectiveness at the willingness-to-pay threshold ranged from £0 to £150,000 was tested by utilizing cost-effectiveness acceptability curves (CEAC).

2.8. Scenario analysis

In this study, we conducted scenario analyses considering uncertainties in model structure and parameters, such as uncertainty in survival data extrapolation, patient medication adherence, medication patterns, and in medication duration, and heterogeneity of utility values.

(1) In scenario 1, we only considered standard parametric survival models.
(2) In scenario 2, the utility values of PFS and PD states reported in the IMpover110 trial (PFS: 0.76; PD, 0.69) were utilized.
(3) In scenario 3, dosing intensity for both atezolizumab and chemotherapy was assumed to be the same as the IMpover110 trial (35).
(4) In scenario 4, it is assumed that patients take vinorelbine orally.
(5) In scenario 5, active treatment during the PD state persisted until 3 months prior to death.
(6) In scenario 6, we adjusted the utilization ratio of vinorelbine or gemcitabine within the range of 0–100%.

2.9. Subgroup analysis

The ICER, probability of being cost-effective at the selected willingness-to-pay threshold, and cost of being cost-effective at the chosen willingness-to-pay threshold for atezolizumab in each subgroup were calculated using subgroup-specific hazard ratios (HRs) of PFS and OS based on Cox proportional hazards models. We considered the subgroup factors of age (≥80, 70–79, or < 70), sex (male or female), race (white or Asian), ECOG PS score (0–1, 2, or 3), tobacco use history (previous, current, or never), histology (non-squamous or squamous), stage (IIIB or IV), brain or liver metastases (yes or no), number of metastatic sites (≥3 or < 3) and PD-L1 expression level (<1, 1–49%, or > 50%).

2.10. Price simulation

The price simulation analysis incorporated fluctuating prices ranging from £1,000 to £3,800, with increments of £10, as per the results from our base-case analysis. Furthermore, Monte Carlo simulation of 1,000 iterations were performed to conduct PSAs for each respective price.

The values, ranges, and sources for all parameters utilized in this model are summarized in Supplementary Table S12.

3. Results

3.1. Model validation

The model's face validity, encompassing its structure, assumptions, data sources, and results, underwent evaluation by clinical experts. The validation results demonstrated a strong fit of our model, as the survival rates for both PFS and OS were consistent with the original data obtained from the IPSOS trial (Supplementary Figure S2). 4-year OS or PFS rates of both atezolizumab and chemotherapy were less than 10%, indicated little uncertainty regards extrapolation (14).

3.2. Base-case analysis results

To conclude, Atezolizumab is not an economical option for patients with advanced NSCLC ineligible for treatment with a platinum-containing regimen as compared to chemotherapy at the current price of £3807.69/1,200 mg. Atezolizumab can be deemed cost-effective only when priced below £2215/1,200 mg at the willingness-to-pay threshold of £50,000/QALY.

The findings of the base-case analysis are outlined in Table 1. The lifetime costs for atezolizumab and chemotherapy amounted to £56,950 and £30,744, respectively. Atezolizumab exhibited a gain of 0.46 life-years and 0.28 QALYs in contrast to chemotherapy. The ICER of atezolizumab compared to chemotherapy were £94,873/QALY, which was higher than the recommended willingness-to-pay threshold of £50,000/QALY, indicating that atezolizumab was not cost-effective when compared to chemotherapy at the current price of £3807.69/1,200 mg. We also conducted an alternative analysis focusing solely on PFS. The ICER for atezolizumab compared to chemotherapy was £213,196/QALY. Breakdown results of costs are provided in Supplementary Table S13.

3.3. Sensitivity analyses

The results of the DSA are shown in Figure 1. The variables that had the greatest impact on the ICER were the price of atezolizumab, percents of patients received subsequent paclitaxel and atezolizumab, discontinuation rate of atezolizumab, and the utility of PD. The price of atezolizumab had the greatest impact on ICER, but in this part, we only considered a limited range of fluctuations, which meant that even at the lowest price, atezolizumab was still not cost-effective. The proportion of patients receiving immune checkpoint inhibitors and other drugs after progression also had a significant impact on ICER, mainly due to the high price of these drugs. Similarly, discontinuation rates related to patient compliance and medication safety were significant factors affecting ICER. Additionally, the impact of utility value on ICER could not be ignored, as it was clearly related to the patient's effectiveness. Lastly, the influence of discount rates was unquestionable, as they were closely related to the results of output and input indicators. Overall, after allowing parameter fluctuated within the specified upper and lower limits, it was established that atezolizumab was unlikely to exhibit cost-effectiveness at the threshold of £50,000/QALY. Nevertheless, this conclusion may be reevaluated if the threshold was set at £90,000/QALY.

The scatter plot and CEAC curve can be found in Figure 2. The results from the PSA showed that the average cost of atezolizumab was £56,943 and the average cost of chemotherapy was £30,754. Moreover, the average effects for these two drugs are 0.86 and 0.58 QALYs, respectively. Following 10,000 iterations, the average ICER is calculated to be £94,384. Atezolizumab was considered to be cost-ineffective at the threshold of £50,000/QALY. Even with a higher threshold of £90,000/QALY, the probability of atezolizumab being cost-effective remained at 40%. The CEAC curve suggested that atezolizumab would be cost-effective if the threshold surpasses £93,700/QALY. Nevertheless, attaining this threshold within the present healthcare landscape in the UK poses significant challenges.

TABLE 1 Results of base-case analysis.

Model	Drug	*Cum* cost (£)	*Cum* life years	*Cum* effect (QALY)	Incremental cost (£)	Incremental effect (QALY)	ICER (£/QALY)
OS	Chemotherapy	30,744	1.07	0.58			
	Atezolizumab	56,950	1.53	0.86	26,206	0.28	94,873
PFS	Chemotherapy	10,325	0.54	0.24			
	Atezolizumab	40,209	0.77	0.38	29,884	0.14	213,196

Cum, cumulative, ICER incremental cost-effectiveness ratio, OS overall survival, PFS progression-free survival, QALY quality-adjusted life year.

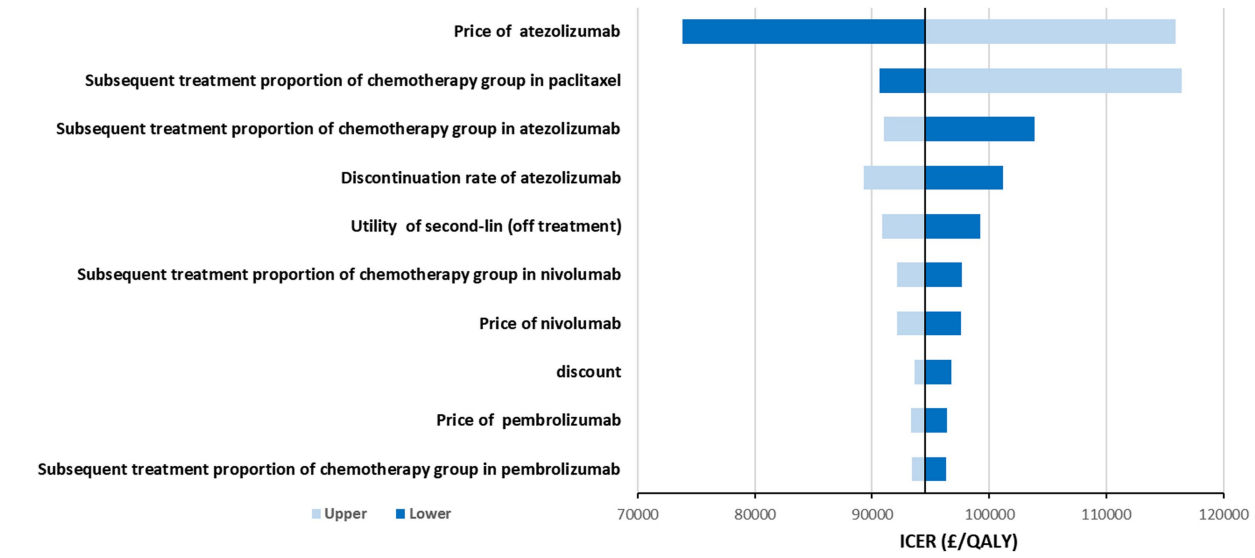

FIGURE 1
Tornado diagram shows the association of variables with the ICER of atezolizumab vs. chemotherapy.

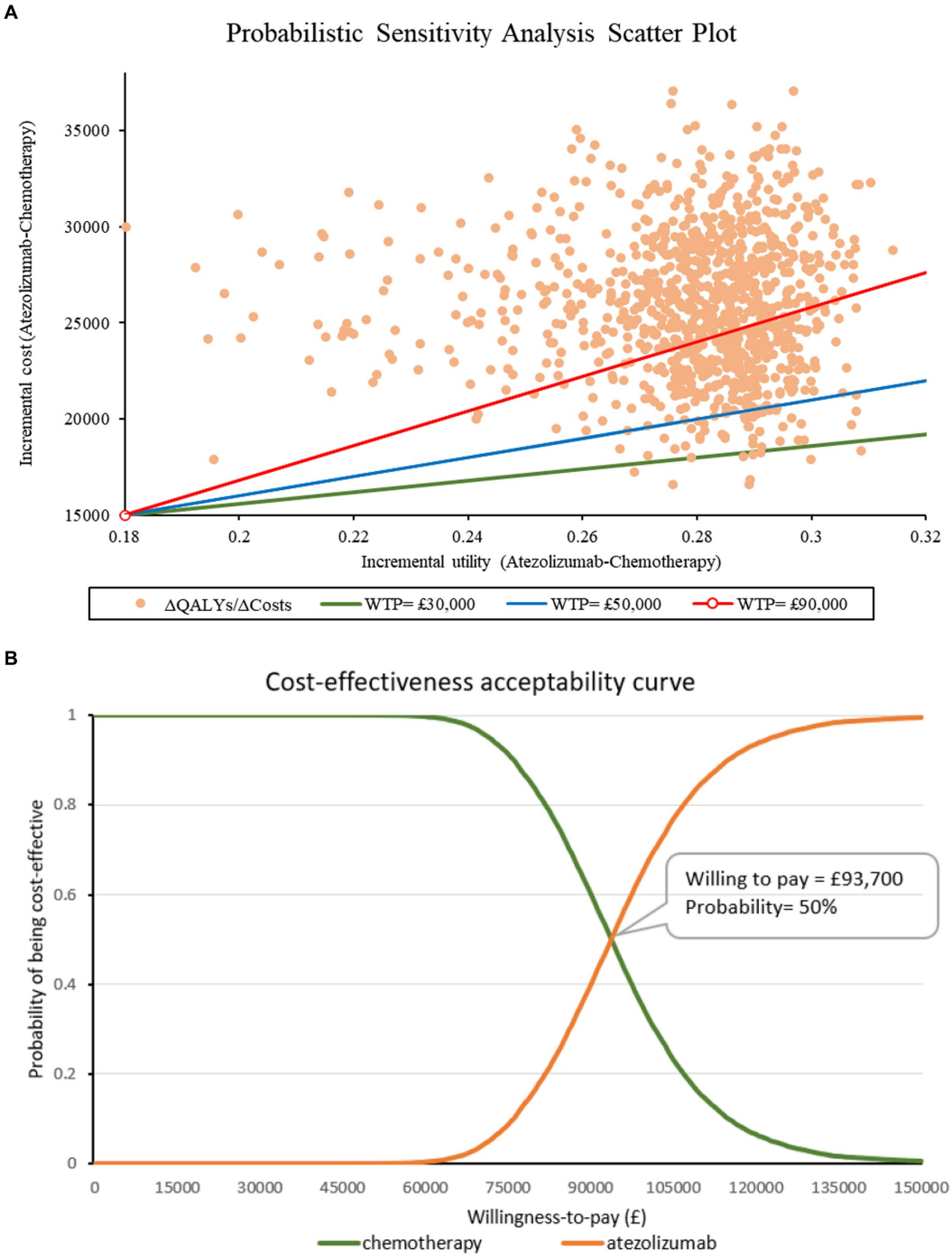

FIGURE 2
Results of the probabilistic sensitivity analysis.

Sensitivity analyses results validated the base-case conclusion. At the threshold of £50,000/QALY, atezolizumab was not cost-effective at the current public price.

3.4. Scenario analysis

In general, the uncertainty related to the structural assumptions and parameter estimates examined had a negligible impact on the base-case conclusion. In scenario 1, utilizing the approaches commonly used that solely incorporate standard parametric models, the findings revealed that the ICER of atezolizumab in comparison to chemotherapy amounted to £99,040/QALY; In scenario 2, employing the utility values documented by the IMpover110 trial, the ICER was £90,974/QALY; In scenario 3, by modifying the dosing intensity for both drugs, the ICER amounted to £88,219/QALY; The ICER of atezolizumab against chemotherapy was £97,354/QALY in scenario 4, where the duration of second-line treatment was altered; In scenario 5, vinorelbine was administered orally, the ICER was £93,335/QALY; In scenario 6, when the usage ratio of gemcitabine ranged from 0 to

100%, the ICER ranged from £92,000 to £98,000/QALY. All scenarios resulted in comparable ICERs and reached the same conclusion. More details, refer to Supplementary Table S14 and Supplementary Figure S3.

3.5. Subgroup analysis

The subgroup analysis results are summarized in Table 2. Overall, the ICERs showed a significant association with HRs, indicating improved outcomes with lower risks of disease progression and death. The ICER for atezolizumab compared to chemotherapy in the entire patient cohort was £120,124/QALY. To meet the cost-effectiveness threshold of £50,000/QALY, the price would need to be £1,970. Atezolizumab did not show favorable results in any of the subgroups at the £50,000/QALY threshold. Atezolizumab was found to be more cost-effective for patients with positive PD-L1 expression (ICER: £72,098/QALY). However, atezolizumab performed worse in patients with PD-L1 expression levels ranging from 1 to 49%, as well as in female patients and those with liver or brain metastases. For other factors, such as age, sex, race, ECOG PS, tobacco use history, histology, number of metastatic sites, and disease stage, did not affect the conclusion, and ICER for atezolizumab compared to chemotherapy in all of these subgroups were over £100,000/QALY. Further details are in Table 2.

3.6. Price simulation

Overall, there is a positive correlation between the cost of atezolizumab and the ICER when compared to chemotherapy. It was concluded that atezolizumab was considered cost-effective if priced below £2,215/1,200 mg, given the willingness-to-pay threshold of £50,000/QALY. Atezolizumab was deemed cost-effective when the price was below £1,465 at a £30,000/QALY threshold. Additionally, for a price of £3,640, atezolizumab was considered cost-effective when the threshold was £90,000/QALY. More findings are depicted in Figure 3 and Table 3. There has been approved Patient Access Scheme for atezolizumab currently in the UK for several indications including advanced NSCLC (17), we believe that implementing the recommended price reductions for atezolizumab for NSCLC patients who are ineligible for treatment with a platinum-containing regimen would be practicality and feasible.

4. Discussion

Atezolizumab was the first to demonstrate a significant enhancement in the survival of patients with advanced lung cancer who have an intolerance to platinum-based chemotherapy (14), irrespective of the type of pathology, PD-L1 expression, or PS score. The real-world NEJ057 study (13) also corroborated this finding. Immunotherapy regimens had significantly higher median OS compared to chemotherapy (19.8 months vs. 9.5 months). Regarding toxicity, findings from both the IPSOS and NEJ057 studies suggested that using immune-mono therapy could significantly decrease the occurrence of severe AEs. Based on the IPSOS trial, it was found that 16% of patients receiving atezolizumab experienced treatment-related severe AEs, while the figure for patients receiving chemotherapy is 33%. Clinical treatment guidelines and strategies is expected to enter a new era of immune-mono therapy. Considering the exorbitant price of atezolizumab compared to the standard treatments, physicians and patients confront the challenge of evaluating its cost-effectiveness. The escalating healthcare costs justify concerns regarding value-based oncology.

This study aimed to fulfill the unmet need for an economic assessment of this novel indication. According to our analysis, atezolizumab was found to be less favorable compared to chemotherapy. When considering a WTP threshold of £50,000/QALY over a lifetime time horizon, atezolizumab incurred an extra cost of £26,206 and had an additional effect of 0.28 QALYs. This led to an ICER of £94,873/QALY in comparison to chemotherapy. Considering parameters' uncertainty, we used the 95% CI as their range of variation. For parameters with only standard deviation or errors, we assume to calculate a 95% CI based on their distribution. For parameters with standard deviation or confidence interval, we assume they vary within a range of ±20%. The findings from the sensitivity analyses provided evidence that the results of the base-case analysis were generally stable and reliable. The factors that had the greatest impacts on economic outcomes were the price of atezolizumab, second-line medication, discontinuation rate of atezolizumab, and utility for PD. To address the uncertainties around the structural assumptions and parameter estimates, multiple scenario analyses were conducted. The selection of survival models, dosing patterns, utility values, and other factors minimally influenced the base-case results and yielded consistent conclusions. Nevertheless, for patients with positive PD-L1 expression, atezolizumab performed better, with an ICER of £72,079/QALY compared to chemotherapy. While for patients with a PD-L1 expression level of 1–49%, female patients, and those with liver or brain metastases, atezolizumab could be cost-effective only when its unit cost lower than chemotherapy. The high diversity of subgroup results reminds us that patient characteristics during the administration of drugs in clinical practice is crucial for utilizing healthcare resources in a rational manner.

Based on the results presented above, price simulations were conducted to explore suitable pricing for atezolizumab for studied patients. It was found that atezolizumab would be cost-efective at a price of £2,215/1,200 mg (a reduction of 41.8%) at the threshold of £50,000/QALY. For patients with positive PD-L1 expression, atezolizumab would be cost-efective at a price of £2,625/1,200 mg (a reduction of 31.1%). At the threshold of £90,000/QALY, atezolizumab would be cost-effective after a 4.4% price reduction for overall patients, and atezolizumab was economical at the current price for PD-L1 positive patients. The benefit of atezolizumab in PD-L1 positive patients was also observed in those with stage II-IIIA NSCLC in the adjuvant setting (4) and those with metastatic NSCLC receiving first- or second-line treatment (36, 37). Nevertheless, atezolizumab was deemed less cost-effective for patients with liver or brain metastases.

The direct and indirect costs associated with advanced NSCLC, particularly as the disease progresses, place a significant financial burden on healthcare systems, society, patients, and caregivers. Therefore, it is of utmost importance to develop newer, more economical, and safer treatments for NSCLC that can effectively slow down or halt the progression of the disease. The well-known fact is that chemotherapy has limited practicality in advanced NSCLC patients, especially in those who are ineligible for treatment with a platinum-containing regimen. Our findings suggest that atezolizumab seems to

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TABLE 2 Summary results for subgroup analyses.

Subgroup	ICER (£/QALY)	Price of atezolizumab[ab] (£) being CE at a WTP of 30,000	Price of atezolizumab[ab] (£) being CE at a WTP of 50,000	Price of atezolizumab[ab] (£) being CE at a WTP of 90,000	Probability of atezolizumab[ab] being CE at a WTP of 30,000	Probability of atezolizumab[ab] being CE at a WTP of 50,000	Probability of atezolizumab[ab] being CE at a WTP of 90,000
All patients	120,124	1,440	1970	3,015	0	0.003	0.229
Age							
≥80	108,555	1,135	2,110	3,265	0.001	0.1	0.388
70–80	306,696	1,210	1,390	1725	0	0.001	0.071
<70	238,188	1,275	1,520	2010	0	0.02	0.177
Sex							
Male	123,542	1,270	1860	2,925	0	0.002	0.225
Female	D	NA	NA	NA	NA	NA	NA
Race							
White	311,108	1,360	1,530	1875	0	0	0.069
Asian	120,995	2,965	1875	2,875	0	0.057	0.349
ECOG PS							
0/1	125,307	1,250	1810	2,850	0.005	0.093	0.328
2	251,359	1,360	1,585	2025	0	0.003	0.074
3	397,937	1,245	1,380	1,660	0.021	0.131	0.283
Stage							
IIIB	98,055	1,335	2050	3,520	0.002	0.152	0.433
IV	205,944	1,340	1,610	2,175	0	0	0.074
Tobacco use history							
Previous	187,340	1,360	1,665	2,295	0	0.002	0.114
Current	110,272	1,280	1910	3,165	0	0.102	0.388
Never	367,872	1,360	1,320	1,215	0.006	0.071	0.222
Histology							
Non-squamous	215,465	1,290	1,565	2,115	0	0	0.119
Squamous	136,042	1,365	1820	2,745	0	0.015	0.248
Brain metastases							
Yes	D	NA	NA	NA	NA	NA	NA
No	163,656	1,325	1700	2,450	0.025	0.099	0.22
Liver metastases							
Yes	D	NA	NA	NA	NA	NA	NA
No	127,801	1,350	1850	2,860	0	0.005	0.218
Number of metastatic sites							
<3	126,383	1,325	1850	2,865	0	0.012	0.281
≥3	337,969	1,280	1,450	1770	0	0.004	0.096
PD-L1 expression level							
<1%	293,190	1,310	1,500	1885	0	0.011	0.13
1–49%	D	NA	NA	NA	NA	NA	NA
≥50%	72,098	1,565	2,625	4,770	0	0.23	0.611

[a]The unit is £/1,200 mg.
[b]with a probability of 50% to be cost-effective.
CE, cost-effective; D, dominated; WTP willingness to pay.

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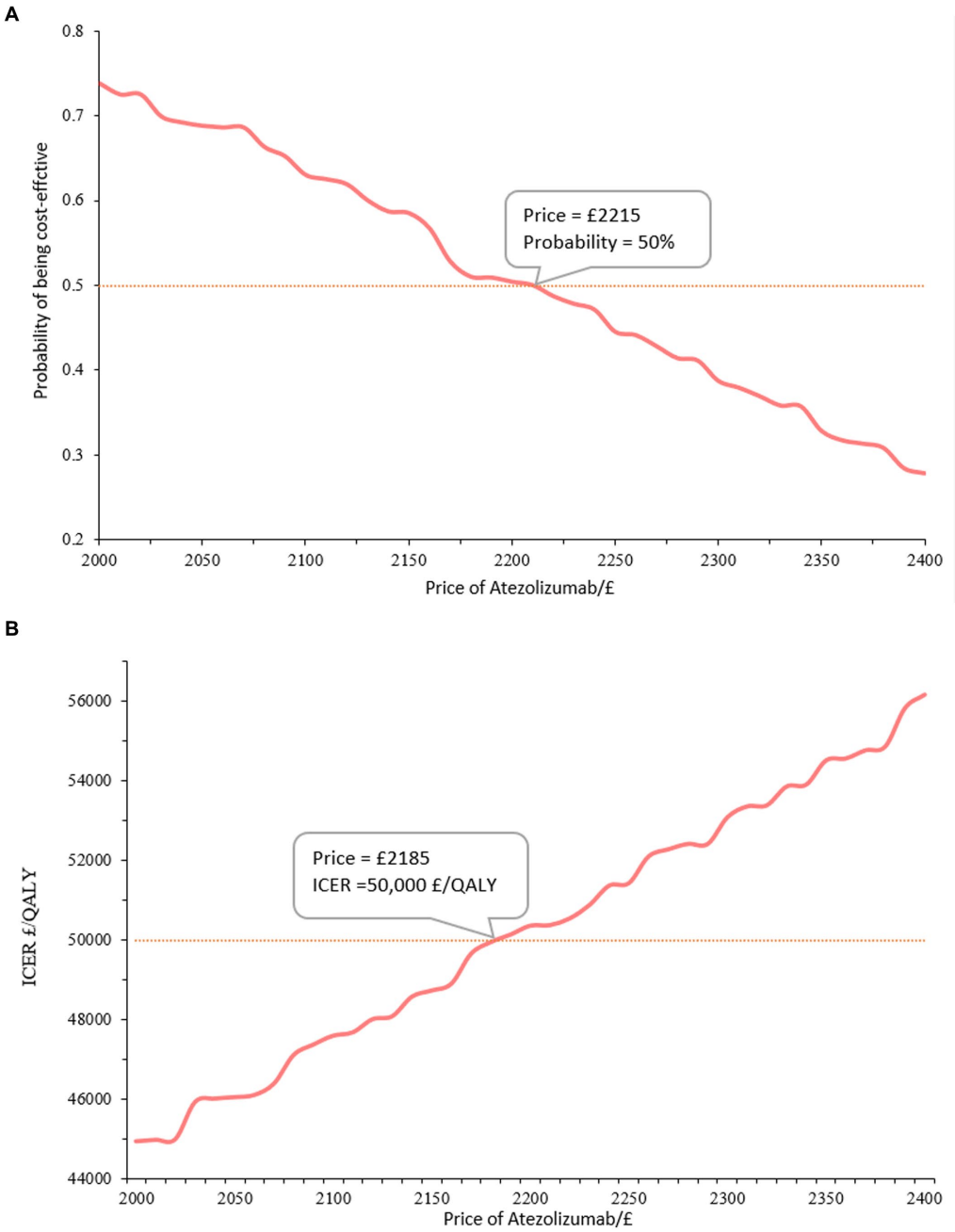

FIGURE 3
Main results of price simulation.

be a potential choice. However, the current price is a deterrent to it becoming the standard of care (SOC). Therefore, price concession is necessary, and our research provide reference for decision-makers. Overall, this research contributes to the development of a new SOC for patients with NSCLC who are unable to tolerate the AEs of the most powerful treatments, and to expedite the approval of this SOC in the UK, thereby providing new treatment options for patients and helping to alleviate the economic burden associated with the disease. Through this study, we aim to inform the UK decision-makers that atezolizumab has the potential to be a new SOC for NSCLC patients who are ineligible for treatment with a platinum-containing regimen. However, at current pricing, it is not yet a cost-effective choice. Furthermore, atezolizumab can be deemed cost-effective only when priced below £2215/1,200 mg at the willingness-to-pay threshold of £50,000/QALY.

The model structure and approach employed in this study are in line with the NICE appraisal of atezolizumab monotherapy for untreated PD-L1 positive metastatic NSCLC (36). For accuracy, reliable sources of information such as NHS reference costs, the PSSRU, and eMIT were utilized. The strengths of this study lie primarily in the innovative research topic, high-quality clinical

TABLE 3 Willingness-to-pay threshold of atezolizumab being cost-effective at specific price.

Price of atezolizumab[a] (£)	WTP[b] (£/QALY)	Price of atezolizumab[a] (£)	WTP[b] (£/QALY)	Price of atezolizumab[a] (£)	WTP[b] (£/QALY)
3,800	94,660	2,800	66,992	1800	39,324
3,700	91,893	2,700	64,225	1700	36,557
3,600	89,126	2,600	61,458	1,600	33,790
3,500	86,359	2,500	58,691	1,500	31,023
3,400	83,593	2,400	55,924	1,400	28,256
3,300	80,826	2,300	53,158	1,300	25,490
3,200	78,059	**2,200**	**50,391**	1,200	22,723
3,100	75,292	**2,100**	**47,624**	1,100	19,956
3,000	72,525	2000	44,857	1,000	17,189
2,900	69,759	1900	42,090		

[a]The unit is £/1,200 mg.
[b]With a probability of 50% to be cost-effective.
WTP willingness to pay.

data, the consideration of various scenarios, and the extensive sensitivity analysis. As far as we know, no other analysis has evaluated the cost-effectivenes of atezolizumab for patients with advanced NSCLC who cannot receive a platinum-containing regimen. Additionally, we performed price simulations to offer decision-makers a more comprehensive comprehension of the economic value attached to atezolizumab. Further discussion is necessary due to the implications of this study. Decision-makers in the UK should be informed about the price at which atezolizumab would be deemed cost-effective for patients with advanced NSCLC who cannot receive treatment containing platinum. Furthermore, our evidence may support the UK health technology assessment submissions for this indication. Finally, our analysis explored the cost-effectiveness outcomes of the 28 prespecified subgroups in the IPSOS trial. NSCLC is in the era of precision treatment (38), economic information for the subgroups may assist in tailoring treatment choices.

Our study has several limitations. First, lack of individual data compelling us to make assumptions about proportional hazards in subgroup analyses. This may cause bias in the calculation of survival rate, thereby leading to errors in the results of subgroup analyses; Second, omitting grade 1 or 2 AEs may have introduced biases, causing the actual cost of treatments to be underestimated. Nevertheless, the sensitivity analysis results indicated that this limitation had minimal impact. Third, at this stage, we did not study the availability and affordability, implying that further research is required. Fourth, as the lack of report in the IPSOS trial (14), the incidences of all grade 3–5 AEs were unavailable. Instead, we could solely consider AEs of any grade with an incidence difference exceeding 5% between groups. This theoretically might result in the underestimation of costs. We contend that its impact was restricted given that the grade 3–5 AEs integrated into our model closely resembled those in the IPSOS trial (14). Our model included 83 events, whereas the IPSOS trial documented 84 events. Fifth, EQ-5D based utility was not collected in the IPSOS trial, and the varying information used for the utility values from different trials might have an influence on the outcomes. Despite conducting a scenario analysis, the impact is still uncertain.

5. Conclusion

From the perspective of the UK healthcare system, atezolizumab is not an economical option for patients with advanced NSCLC ineligible for treatment with a platinum-containing regimen. Moreover, atezolizumab can be deemed cost-effective only when priced below £2215/1,200 mg at the willingness-to-pay threshold of £50,000 per QALY. Our study may offer evidence to guide the assessment of therapeutic alternatives and pricing setting for advanced NSCLC.

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 authors.

Author contributions

YJ: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. MZ: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. JX: Data curation, Supervision, Writing – review & editing. JL: Investigation, Methodology, Writing – review & editing. WT: Supervision, Writing – review & editing, Funding acquisition, Resources, Validation, Visualization. XZ: Investigation, Methodology, Supervision, Validation, Visualization, Writing – review & editing.

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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.

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Supplementary material

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Micro-costing analysis of suspected lower respiratory tract infection care in a French emergency department

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Introduction: In the context of budgetary constraints faced by healthcare systems, the medical-economic evaluation of care strategies becomes essential. In particular, valuing consumed resources in the overcrowded emergency departments (EDs) has become a priority to adopt more efficient approaches in treating the growing number of patients. However, precisely measuring the cost of care is challenging. While bottom-up micro-costing is considered the gold standard, its practical application remains limited.

Objective: The objective was to accurately estimate the ED care cost for patients consulting in a French ED for suspected lower respiratory tract infection.

Methods: The authors conducted a cost analysis using a bottom-up micro-costing method. Patients were prospectively included between January 1, and March 31, 2023. The primary endpoint was the mean cost of ED care. Resources consumed were collected using direct observation method and cost data were obtained from information available at Strasbourg University Hospital.

Results: The mean cost of ED care was €411.68 (SD = 174.49). The cost elements that made the greatest contribution to the total cost were laboratory tests, labor, latency time, imaging and consumables. Considering this cost and the current epidemiological data on respiratory infections in France, the absence of valuation for outpatient care represents an annual loss of over 17 million euros for healthcare facilities.

Conclusion: Micro-costing is a key element in valuing healthcare costs. The importance of accurately measuring costs, along with measuring the health outcomes of a defined care pathway, is to enhance the relevance of health economic evaluations and thus ensure efficient care.

KEYWORDS

cost analysis, emergency department, respiratory infections, health economics, micro-costing

1. Introduction

In recent years, emergency departments (EDs) have faced major challenges related to overcrowding. In the current ED situation, medico-economic assessments are needed to optimize resource allocation without compromising the quality of care for a growing number of patients (1, 2). Indeed, to make informed healthcare decisions, it is essential to compare both the costs and healthcare outcomes associated with the interventions under consideration (3). Healthcare decision-makers therefore need to identify which processes can be improved, at the right cost, and how this cost relates to health outcomes (4, 5). The accuracy of cost estimates is therefore of paramount importance, as it conditions the evaluation of the effectiveness of strategies. However, estimates of patient care costs are often imprecise and based on overall hospitalization costs, making it difficult to assess the cost of specific emergency care.

It is usual to consider that the choice of costing method involves a balance between accuracy and implementation feasibility (5–7). Costing methods are usually divided into four categories, depending on the monetary valuation of resources (bottom-up versus top-down), and the precision of the measurement (micro-costing versus gross-costing) (6, 8). The micro-costing methods are the most precise costing methods that involve direct, detailed, real-time observation of the resources consumed at each step of each patient's care (7). It enables the monetization of resources consumption observed at an individual level, thereby facilitating the calculation of a per patient cost of care which will be used to estimate the average cost of the intervention studied. This stands in contrast to the top-down method, which assigns each patient an average cost based on aggregated data (8). While the bottom-up micro-costing method is considered the gold standard for hospital cost estimation due to its precision (3), its practical implementation is often limited due to a significant consumption of labor time and resources (6, 7). The need for detailed data collection at the patient level can be time-consuming and costly, making it less feasible for large-scale studies or healthcare facilities with limited resources. Therefore, despite its accuracy, practical constraints often lead researchers to explore alternative costing methods, such as gross-costing or top-down approaches.

The valuation of healthcare resources consumed is increasingly important in the context of EDs experiencing misuse and overuse. In France, the cost paid to hospitals for outpatient care is based on health insurance reimbursement data, and does not take into account the resources actually consumed during treatment, which certainly underestimates the cost. The extreme situation of the COVID-19 pandemic and the associated hospital saturation, highlighted the importance of setting efficient care pathways within a short time and to rethink care pathways (9, 10). This analysis can be extended to the broader context of respiratory infections ED care. Nevertheless, there is a gap in knowledge when it comes to estimating costs and determining the optimal care pathway for ED outpatients (11). To ensure an accurate cost estimation, a recommended set of steps was proposed (4, 5), guiding the process from selecting a pathology or symptom and defining the patient's care pathway to calculating the total cost of patient care by estimating the time and cost associated with each step. Based on these recommendations, the main objective of the present study was to accurately estimate the total cost associated with the management of patients presenting with suspected lower respiratory tract infection, a frequent reason for ED visit, in the emergency department of Strasbourg University Hospital (France) by a bottom-up micro-costing method. In this regard, the feasibility of implementing a bottom-up micro-costing technique to calculate the ED care costs has also been assessed. Secondary objectives included identifying the care pathway and cost elements associated with ED care for these patients and assessing the potential financial impact of micro-costing-based ED care cost estimation on healthcare system expenses.

2. Methods

2.1. Study design, setting, and population

First, the authors conducted a cost analysis using a bottom-up micro-costing method. The primary endpoint was the mean total cost of ED care. A set of recommended steps was used to ensure an accurate cost estimation (4, 5). Thirty patients with a suspected lower respiratory tract infection who visited the ED during business hours were prospectively included between January 1, 2023 and March 31, 2023. To ensure the best representation during this study period, patients were systematically and consecutively included when they arrived at the ED with clinical or historical criteria compatible with a potential lower respiratory tract infection. These criteria included a patient's history of symptoms such as cough, sputum production, purulent secretions from the airways, shortness of breath, fever when no other infectious cause besides respiratory was initially suspected, chest pain in the absence of other factors suggesting a non-pulmonary origin, a history of asthma or exacerbation of chronic obstructive pulmonary disease, contact with infected individuals, and referral to the ED by a healthcare professional for suspected lower respiratory infection, either based on clinical symptoms or the results of prior complementary examinations conducted before ED admission.

Then, a budget impact analysis was conducted using the findings from this study and data from the French Health Insurance reimbursement tariffs.

2.2. Identification of resources

Before conducting the cost analysis, on-site observations were performed to determine the relevant cost elements to be considered. The patient's clinical pathway steps and resources used during ED care were established in consultation with two expert physicians, validated during on-site observations, and refined iteratively during the cost analysis. The clinical pathway within the Strasbourg ED is presented in Supplementary Figure S1.

2.3. Data collection: measurement of resources

The duration of procedures, consumables used and additional investigations were measured through direct observation from patient's arrival until the medical decision regarding orientation (discharge or hospitalization). The resources used from the medical decision regarding orientation until effective hospitalization or

discharge (referred to as latency time) were estimated based on the patient's electronic medical records.

2.4. Valuation of resources

To ensure maximum accuracy, the valuation of resources was based on local unit costs (12). This included staff salaries, the purchase price of drugs, the purchase price of all consumables, and the laboratory's invoiced price for biological analyses.

The cost of labor was calculated by multiplying the observed time spent on a task (in minutes) by the cost of the human resource(s) involved (in euros/min).

The cost of consumables and laboratory tests was calculated by multiplying the unit cost (in euros) by the number of units consumed.

Since the scanner installed in the ED had already been fully amortized, its utilization had minimal influence on the ED care cost. Therefore, the cost of imaging was estimated for each patient based on the reimbursement tariffs set by the French Health Insurance provided below:

- Chest X-ray: €21.28
- Chest CT scan: €58.77
- Chest CT-scan with contrast: €68.37
- Thorax, abdomen and pelvis CT scan: €84.04
- Thorax, abdomen and pelvis CT scan with contrast: €93.64

The cost of a CT scan includes the procedure fee (€32.00), the technical flat rate (€25.27 for chest CT scan and €50.54 for thorax, abdomen and pelvis CT scan) and the storage cost (€1.50). The use of contrast incurs an additional cost of 9.60€ to which the cost of consumables used for injection must be added.

The cost of equipment was considered negligible therefore not included in the total cost.

All cost elements were integrated into an Excel spreadsheet, allowing for the calculation of the total cost per patient (Supplementary Table S1).

2.5. Financial valuation of latency time

The latency time, as referred to in this study, represents the time between the medical decision to hospitalize or discharge a patient, and the patient's transfer from the ED to the inpatient department or return home.

Based on the patient electronic medical record, the following costs were taken into account for the monetary valuation of resources consumed during the latency time:

- additional laboratory tests conducted,
- consumables and labor required for venipuncture and/or arterial blood gas, as measured during the direct observation process,
- nurse and nursing assistant labor per 8-h shift, using the mean duration of bedside care as observed during the direct observation,
- medical examination per 12-h shift, using the mean duration of clinical examination as measured during the direct observation.

2.6. Statistical analysis

At present, there are no specific guidelines regarding the number of patients to include or the minimum level of precision required in micro-costing studies (8). To the best of the authors' knowledge, there is no micro-costing study in the literature focusing on the costs of ED care for specific clinical pathways. In this study, the authors drew upon existing research conducted using a cost calculation method similar to micro-costing known as Time Driven Activity-Based Costing (TDABC). The decision was made to include 30 patients, aligning with the average number of patients or clinical pathways studied in TDABC research conducted in EDs, which ranges from 8 to 113 procedures (13–16). Traditional frequentist statistical methods cannot estimate the precision of the mean cost calculated through micro-costing, due to absence of existing data in the literature regarding the variance of this cost. The use of Bayesian statistical methods allows for the establishment of a maximum credibility interval range of approximately €500 around the calculated mean when 30 patients are included. This approach relied on estimates of minimum and maximum cost values provided by two expert physicians.

All statistical analyses were conducted using the 4.2.1 version of R statistical software. The quantitative variables were described using the mean, along with the corresponding standard deviation, minimum, and maximum values.

A multiple linear regression was conducted to examine the relationship between the total ED care cost and the various cost elements considered in this study. Assumptions concerning linearity, independence, and distribution were verified. Goodness-of-fit statistics, including BIC and adjusted R-squared, were examined to demonstrate that the selected model was the best fit for the data among the tested models.

A linear regression model was employed to determine the presence of a significant linear association between the total cost and the length of stay in the ED. Assumptions of linear regression were assessed to ensure the validity of the model.

3. Results

3.1. Care process map

Direct observation and time measurement were conducted throughout the care pathway for suspected lower respiratory tract infection in the emergency department (ED). The process map, illustrating the various steps and their corresponding durations is represented in Figure 1. The mean duration of ED care prior to the orientation decision was 7 h and 23 min (SD = 3 h and 30 min).

Once the orientation decision was made by the physician, the patient waited an average of more than double that duration (16 h and 5 min, SD = 21 h and 22 min), whether it was for hospitalization or discharge after monitoring and clinical reassessment. The average duration of each step is presented in Table 1.

3.2. Cost elements

The mean total cost of ED care for suspected lower respiratory tract infection, estimated using a bottom-up micro-costing method,

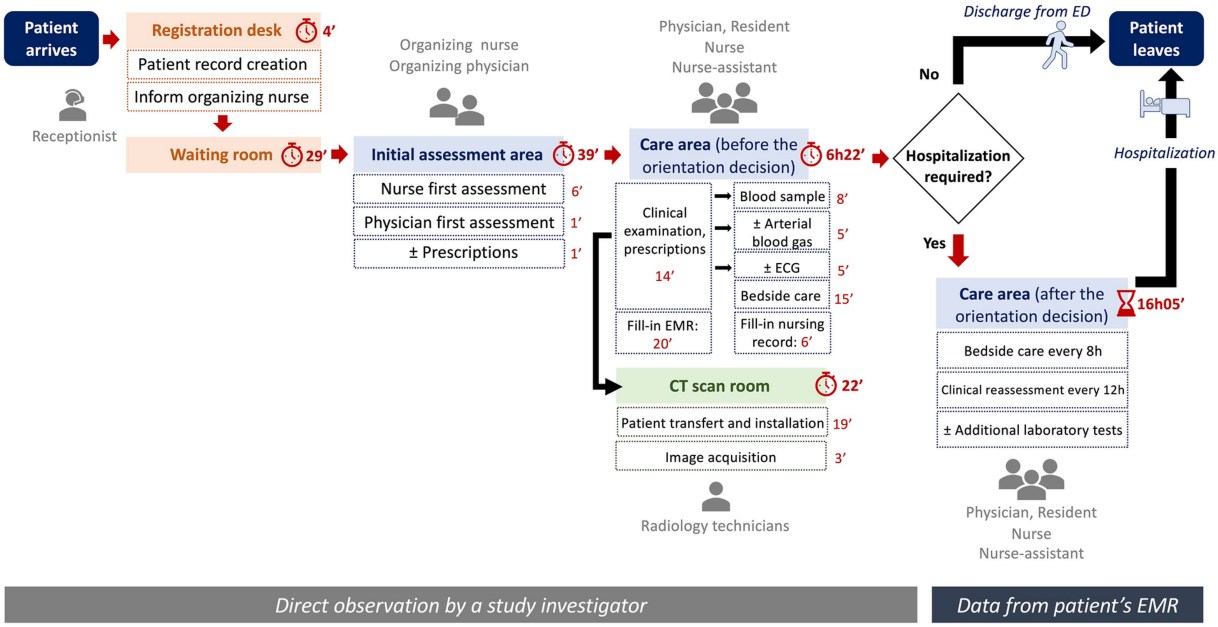

FIGURE 1
Care process map for a suspected lower respiratory tract infection in the ED. ED, emergency department; min, minutes; h, hours; EMR, electronic medical record; ECG, electrocardiogram.

TABLE 1 Mean duration for each step.

Steps	Mean duration	Minimum duration	Maximum duration	Standard deviation
Waiting room	29 min	1 min	1 h44	35 min
Initial assessment area	39 min	6 min	1 h44	36 min
Nurse first assessment	6 min	2 min	24 min	5 min
Physician first assessment	1 min	1 min	5 min	2 min
Care area	6 h22	1 h16	13 h44	3 h33
Clinical examination	10 min	3 min	25 min	5 min
Bedside care	5 min	1 min	20 min	6 min
Blood sample	8 min	2 min	50 min	9 min
Arterial blood gas	5 min	1 min	25 min	5 min
Monitoring vital signs	5 min	1 min	24 min	5 min
CT scan room	22 min	8 min	1 h34	19 min
Duration of ED care	7 h23	2 h53	18 h19	3 h30

min, minutes; h, hours; CT, computed tomography; ED, emergency department.

is €411.68 euros (SD = 174.49). The cost varied from a minimum of €167.96 to a maximum of €1033.18. The mean cost for each resource category is presented in Table 2.

The most important cost elements were laboratory tests, accounting for 37.8% of the total cost, followed by labor (22.1%), latency time (17.2%), imaging (14.7%) and consumables (7.2%). The cost of medication contributed the least to total cost (1.1%).

To illustrate the relationship between the total cost of ED care (dependent variable, Y) and the cost elements (independent variables, X_i), a descending stepwise procedure was employed to select the best-fitting model after removing multicollinear variables. The resulting multiple regression model, $Y = 180.98 + 0.98 \times_1 + 1.09 \times_2$, where X_1 and X_2 represent the cost of laboratory tests and latency time, respectively, exhibited an R-squared value of 0.92. Notably, despite regression coefficients close to 1, there was no evidence of collinearity between the two independent variables (VIF < 1.5). This analysis suggests that 92% of the cost variability observed in this study can be attributed to the costs of laboratory tests and the costs associated with latency time.

3.3. Relationship between the cost and the length of stay in the ED

The linear regression model identified a significant linear relationship between the total cost of ED care and the length of stay (LOS) in the ED ($\beta = 0.11$, $p < 0.001$), with a coefficient of

TABLE 2 Mean cost by type of resource (in euros, €).

Type of resource	Mean cost	Standard deviation
Labor	€90.78	30.82
Receptionist	€1.83	0.64
Paramedical team	€42.87	15.82
Nursing assistant	€3.40	2.56
Nurse	€31.37	13.98
Radiology technician	€8.10	3.93
Medical team	€45.82	20.23
Emergency resident	€24.24	15.42
Emergency physician	€21.58	16.76
Laboratory tests	€155.72	85.57
Blood tests	€87.83	28.32
Microbiological analyses	€67.89	81.78
Consumables	€29.47	21.89
Blood sampling equipment	€4.80	6.03
Microbiological sampling equipment	€18.14	19.14
Wound dressing equipment	€0.11	0.09
Infusion equipment	€1.44	0.96
Oxygenation equipment	€2.21	6.12
Hygiene equipment	€0.62	0.46
Other equipment	€1.31	0.85
Stationery	€0.18	0.03
Imaging	€60.53	25.14
Medication	€4.51	6.02
Latency time cost	€70.67	114.58
Total cost of emergency department care	€411.68	174.49

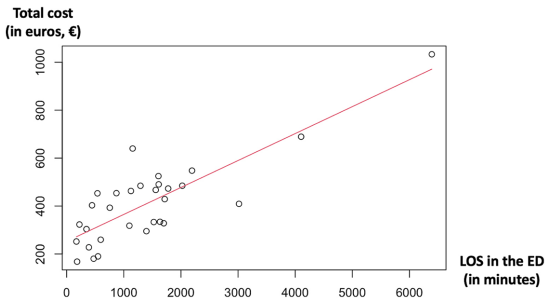

FIGURE 2
Total cost of ED care versus length of stay in the ED. The line corresponds to the linear regression line. LOS, length of stay; ED, emergency department.

determination (R-squared) of 68%. These findings suggested that higher LOS in the ED were associated with higher costs. The patient with the longest LOS, specifically 4 days, 10 h, and 31 min, incurred the maximum cost of €1033.18. The distribution of total costs versus LOS is presented in Figure 2.

3.4. Budget impact analysis

Each year in France, approximately 500,000 patients are diagnosed with lower respiratory tract infections. Among them, 180,000 will seek care at the ED, and 64% of them require hospitalization. Thus, over 65,000 patients per year are treated as outpatients (17). There is no consensus on the financial assessment of ED care for outpatients, and consequently, no available data on this matter. However, for hospitalized patients, ED care costs are standardized at €150, as part of the Diagnosis-Related Group, calculated using gross-costing methods and used for reimbursement by the French Health Insurance. Subtracting this €150 ED care cost from the one obtained through micro-costing in the present study reveals an additional cost of €261.68. By extrapolating this cost to the 65,000 yearly outpatients, the hospital incurs an annual financial loss exceeding €17,000,000.

4. Discussion

This study marks a significant step in healthcare cost analysis, as it is the first comprehensive bottom-up micro-costing study to assess the entirety of care costs associated with patients visiting a French ED for suspected lower respiratory tract infections. In a context where specific data on ED care costs in France are lacking, this research fills a significant gap by providing an initial, highly precise estimate of these costs through the most rigorous calculation method available. Importantly, the successful implementation of this micro-costing approach underscores its feasibility for similar investigations, opening doors to more detailed cost analyses in healthcare settings. However, it's worth noting that a very small portion of the total cost could not be calculated with the bottom-up micro-costing technique due to equipment depreciation. This situation, which may also arise in other healthcare facilities, leads to considering the development and evaluation of hybrid cost calculation approaches in future studies. While the assumptions made likely have a minimal impact on the overall cost, they should be considered when evaluating the accuracy of the cost estimates.

In addition to its originality, this study reveals how costs elements are distributed throughout the ED care process, offering valuable insights for future resource allocation or, where appropriate, process improvement. It seems that acting on ED latency times and on the prescription of biological tests could represent significant factors for influencing the total cost of ED care. Indeed, the analysis reveals the substantial impact of latency time as the third most important cost element, indicating the strain on hospital bed capacity. A linear relationship was observed between the LOS in the ED and the mean cost of care. To confirm this association, it would be necessary to consider potential confounding factors that influence both the LOS and costs, such as the patient's condition and the severity of the respiratory infection being treated. These specific data were not collected in this initial exploratory cost-focused study and may be the subject of future research. The budget impact analysis for outpatient costs emphasizes the importance of accurate valuation to ensure appropriate reimbursement for ED services. Given the substantial

potential annual loss of over 17 million euros when employing gross-costing methods instead of a bottom-up micro-costing approach for outpatient care, it becomes imperative to acknowledge the broader implications. Inaccurate cost calculation methods that fail to consider actual healthcare facility expenses can compromise healthcare system sustainability, hinder patient access to quality care, and challenge resource allocation strategies. In a healthcare landscape characterized by escalating costs and growing demands, optimizing cost calculations through micro-costing, when proven appropriate, appears crucial to ensure the long-term viability of healthcare systems and equitable access to care for all.

Several studies have demonstrated significant differences between micro-costing and gross-costing methods, with the latter potentially leading to over – or underestimation of costs (18–20). Bottom-up micro-costing provides precise cost estimates by meticulously tracking every resource used in patient care. It offers high accuracy but demands extensive data collection, making it resource-intensive and potentially less feasible for large-scale studies (6, 8). An alternative approach has shown promise in estimating costs of ED care. This method called Time-Driven Activity-Based Costing (TDABC) simplifies cost estimation by assigning standard costs to activities based on time estimates rather than tracking the precise resource consumption associated with each activity, enhancing feasibility and reducing resource consumption (13–16, 21). However, its accuracy may be lower as it relies on approximations. Both methods seem valuable approaches for healthcare cost analysis. For detailed, resource-rich studies, bottom-up micro-costing excels in accuracy. In contrast, TDABC offers a practical compromise when resource constraints or broader-scale analyses come into play. While there is no definitive consensus on the preferred costing method, evidence from the literature suggests that micro-costing methods should be prioritized whenever feasible. Knowing a more precise cost of ED care could encourage decision-makers to distinguish this cost in order to better value the activity of EDs, especially for outpatients.

4.1. Limitations

The cost analysis was conducted in a single center, based on a limited number of patients. Since this was an initial feasibility study with an exploratory objective, a sample of 30 patients was deemed adequate, considering the limited available data from precise costing methods such as micro-costing in the literature. Despite the systematic and consecutive inclusion process to ensure the best representation, the study period is limited to regular working hours and the first trimester of the year (January to March). The cost range obtained in this study is only applicable to lower respiratory tract infections occurring during this time of the year and may not correspond to other epidemic peaks or patients admitted to the ED during night shifts. It should be noted that the cost estimation was based on specific local unit costs of Strasbourg University Hospital. It is noteworthy that emergency care protocols for suspected lower respiratory tract infections, along with the complementary diagnostic examinations such as imaging and laboratory tests, typically adhere to standardized guidelines and procedures consistent across healthcare facilities in France. As a result, the cost data collected in this study can offer valuable insights with potential applicability to other hospitals in France, given the standardized nature of care practices in this context. Nevertheless, it is essential to acknowledge that despite the protocolization of certain aspects of emergency care, variations in resource utilization, administrative practices, and local factors may still influence cost profiles. Therefore, while these study findings provide interesting insights, it is important to recognize the potential for variations among hospitals. To apply the findings from this study in different contexts and centers, one should consider seasonality and cost data from the studied hospital (unit prices of consumables, laboratory analysis costs, fully loaded staff salaries, and whether or not imaging equipment is depreciated). The cost data for ED care before the decision on patient orientation can reasonably be replicated by following a similar clinical pathway. These represent standard ED care practices in the context of consultations for suspected lower respiratory tract infections in French EDs. However, the time delay before transfer to the hospitalization department can vary depending on the organization and capacity of the center under study. Calculating this part of the total ED care cost would require specific consideration for each individual center.

Despite the apparent feasibility of employing a bottom-up micro-costing method, it is important to acknowledge that not all costs could be calculated using this technique. This approach aligns with the suggestions of Jacobs et al. and Tan et al., who propose the utilization of multiple costing methods within a single study, thereby disaggregating the total cost into individual cost elements and leveraging the strengths of each method while mitigating their weaknesses (7, 22, 23). Limiting the application of bottom-up micro-costing to cost elements that exert a great impact on the total cost still allows for a reliable estimate of the total cost (6).

It would have been valuable to compare the micro-costing estimate of ED care costs from the present study with previous gross-costing estimates. However, no data in the literature are available for such comparisons with results from similar populations or time periods.

5. Conclusion

The study enabled to accurate estimation of the total cost of emergency department (ED) care for patients presenting with suspected lower respiratory tract infection in a French ED, employing a bottom-up micro-costing method.

The study also allowed the identification of the care pathway and cost elements associated with ED care for these patients, and demonstrated the potential financial impact of managing outpatients. If used in policy decisions, these findings could guide cost-effective strategies for managing patients with respiratory infections, improving cost assessment, and optimizing resource allocation. This evidence-based approach could also apply to other common ED visits, contributing to the development of efficient healthcare policies for different conditions.

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 humans were approved by the ethics committee of Strasbourg University Hospital (CE 2021-141). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

VW: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Software, Validation, Visualization, Writing – original draft, Writing – review & editing. É-AS: Methodology, Supervision, Writing – review & editing. PL: Writing – review & editing. ÉB: Investigation, Writing – review & editing. PB: Writing – review & editing. SK: Conceptualization, Methodology, Supervision, Visualization, Writing – review & editing.

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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.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1276373/full#supplementary-material

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Differences of corruption types in selected Western and central-eastern health systems during the COVID-19 pandemic: a rapid review

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Objectives: To identify, describe, and classify the cases of health corruption present in selected Western [the Netherlands and the United Kingdom (UK)] and Central-Eastern European (Poland and Slovakia) countries during the COVID-19 pandemic.

Methods: A rapid review of the literature was conducted, evaluating data from 11 March 2020 to 15 April 2021. Information sources included MEDLINE via WoS, IBSS via ProQuest, Scopus, and gray literature.

Results: Thirteen cases were identified across the four countries. The primary type of health corruption in Western European countries was procurement corruption, while misuse of (high) level positions was the most prevalent in Central-Eastern European countries. Actors from central governments were most involved in cases. The rule of law and anti-corruption watchdogs reported most cases in the United Kingdom and the Netherlands, while the media reported cases in Poland and Slovakia.

Conclusion: The differences in types of corruption in WE and CEE countries emphasize the need to contextualize the approach to tackle corruption. Thus, further research in preventing and tackling corruption is a vital and necessary undertaking despite the inherent of conducting health corruption research.

KEYWORDS

health corruption, COVID-19, public health crisis, Western Europe, Central-Eastern Europe

Introduction

Corruption is the abuse of entrusted power for private gain and has a significant impact on health systems (1). Corruption costs developing countries over one trillion USD every year. Health care systems (HCSs) themselves lose out on over 500 billion USD annually due to the impact of corruption (2). This impact is not limited to monetary losses, as improvements in the control of corruption result in the more efficient use of healthcare resources. This leads to better outcomes, improving the population's overall health (3). For instance, Lio and Lee reported that

just a 1-point difference in the World Bank's Control of Corruption Indicator is associated with numerous positive effects. These include a longer life expectancy (0.44 more years), lower rates of infant mortality (2.67 fewer deaths per 1,000 live births), and lower rates of under-five mortality (4.62 fewer deaths per 1,000 children) (4).

Corruption in health systems remains understudied despite its impact on financial and healthcare outcomes. Around half of the literature that combines the terms 'health' and 'corruption' in PubMed -one of the leading biomedical databases- was produced from 2019 onwards. However, the diversity in terms used to refer to corruption, as well as the lack of mechanisms that protect those reporting corrupt acts, contribute to the difficulty of studying corruption in health systems.

At the same time, corruption tends to increase during periods in which health systems are at their most vulnerable. These include public health crises like the COVID-19 pandemic, in which attention is focused on the response to the crisis. The need for swift action to protect the population compromises the quality of procurement, while modifications in existing processes have unintended consequences on the risk of corrupt practices. This was evident during the COVID-19 pandemic, in which the nature of procurement changed dramatically. For example, reduced scrutiny in the face of necessity led to actors from healthcare systems engaging in corrupt practices (5).

Social and cultural context are additional elements to consider in the study of corruption in health systems. Societies' perception of corruption varies from country to country, imposing challenges in the reporting of corrupt practices. An example of this is the perception of giving gifts to obtain something from public service, with Central and Eastern European (CEE) countries having a higher acceptance of this than Western European (WE) countries (6). The differences in corruption between WE and CEE countries are also evident in corruption performance indicators. Take, for instance, the Control of Corruption Index (measured from 0 to 100 - the higher the number, the better the control of corruption) by the World Development Indicators. This showed that the United Kingdom and the Netherlands scored 94 and 96 respectively, while Poland and Slovakia scored 73 and 66. Similarly, we can look at the rule of law, which can be understood as "the principle that political power must be exercised in accordance with law rather than in an arbitrary or self-interested manner, and that disputes among private individuals and between them and the Sovereign must be subjected to independent adjudication." (7) The Rule of Law Index (measured from 0 to 1 - the closer to zero, the lower the adherence to the rule of law) captures compiles data from nine factors to provide a quantitative measure of this principle (7). According to this measure, it is evidenced that the rule of law is higher in the United Kingdom and the Netherlands (0.82 and 0.88 respectively) when compared to Poland and Slovakia (0.73 and 0.51 respectively) (8).

While there are available reports from CEE and WE countries of corrupt practices during the COVID-19 pandemic, most of these are presented as single cases from local facilities (9), narrative reviews (10), or evaluations of a country's procurement by anti-corruption organizations (5, 11). These reports provide an important contribution to the field. However, there is a need for a systematic evaluation that considers the context of individual countries. This will enable the identification of corrupt practices that might not be perceived as such in other societies.

It is important to contextualize the evaluation of corrupt practices during crises. This arises from the need to create policies that will prevent and promptly identify the most prevalent types of corruption in each individual country. Moreover, the study comes at a time of poly-crises; namely the Russian invasion of Ukraine, the cost-of-living crisis, and the worsening impacts of climate change (12). These factors all highlight the need to ensure that resources are used efficiently to protect the population's health.

This study acknowledged the differences between the perception and the state of corruption in WE and CEE countries, together with the increased vulnerability of health systems to corruption during public health crises. Consequently, the aim was to identify, describe, and classify the cases of health corruption present in selected Western and Central-Eastern European countries during the COVID-19 pandemic.

The vulnerability of health systems and its actors to corruption

Health systems are defined as the actors (organizations, institutions, people) whose primary goal is to improve health (13). In order to achieve this goal, actors engage in interactions both inside and outside the health system (Figure 1). For instance, the COVID-19 pandemic required both the redesign of existing hospitals and the creation of new ones. This involved not only the healthcare sector, but also the construction, financial, and environmental sectors as well.

While health system actors aim to improve health, these interactions between sectors occur under an asymmetric distribution of information, also known as the principal-agent (PA) problem. In context of healthcare, the PA problem refers to the provider (agent) of a service maximizing profits at the expense of the actor in the system (principal) (14). An example of how the PA problem was present during the COVID-19 pandemic is the allocation of financial resources to car manufacturers (*agent*), who falsely claimed to be able to provide ventilators to health systems (*principal*) (15).

In addition to the PA problem, other factors contribute to the occurrence of corrupt practices in health systems. Vian developed a theoretical framework that illustrates how the interactions between health system actors lead to the abuse of entrusted power for private gain (16). The framework considers three main factors that influence actors to engage in corrupt practices:

1. Rationalization.
2. Opportunity to abuse.
3. Pressure to abuse.

Rationalization refers to behavior influenced by social norms and ethical beliefs. The opportunity to abuse is influenced by a country's health system structure, and includes the level of monopoly, discretion, accountability, citizen voice, transparency and enforcement. Lastly, the pressure to abuse stems from pressure from clients, as well as that from wages or incentives. Based on Vian's theoretical framework, we hypothesized that the structural and social differences between WE and CEE countries would yield to different types of corruption during the COVID-19 pandemic.

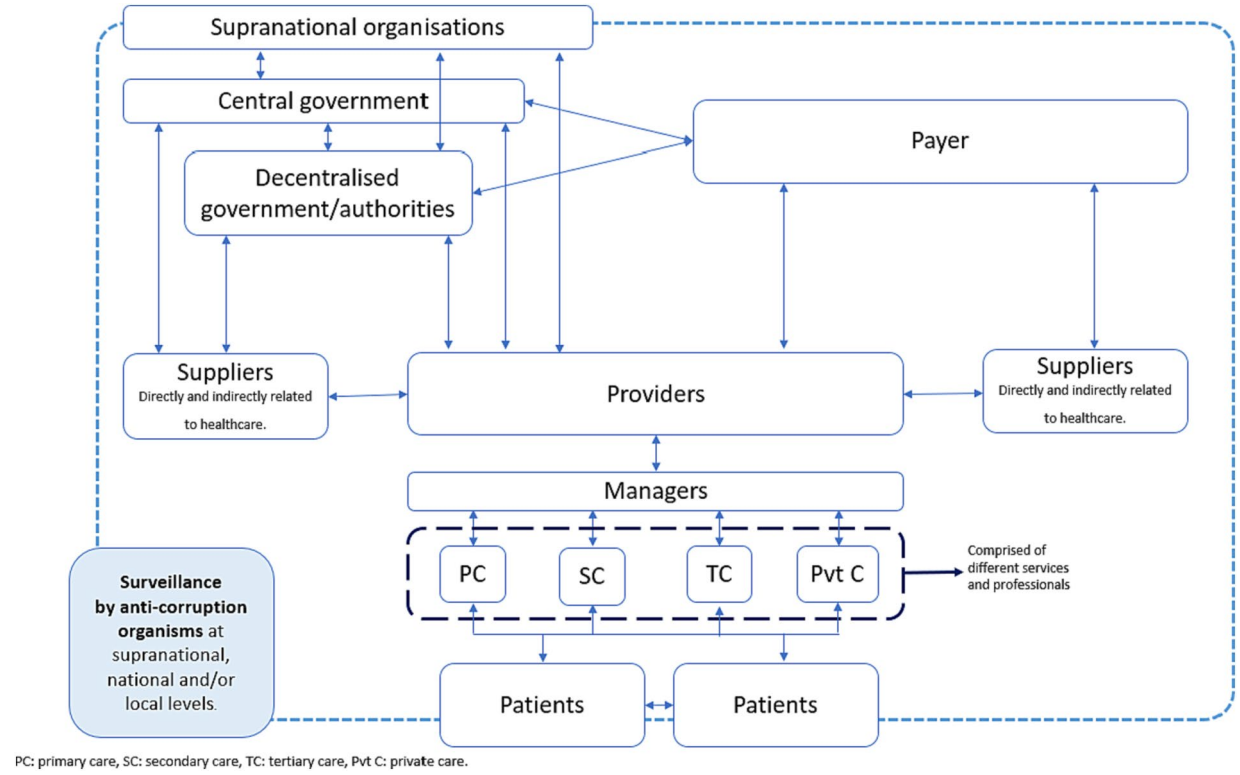

FIGURE 1
Interactions between actors from the health system (8).

Methods

The highly dynamic pace of the pandemic required a fast approach to studying how health corruption unfolded through the different waves. Therefore, a rapid review of the literature was considered the most appropriate approach. This study followed the practical guide for rapid reviews by the World Health Organization (17). An internal protocol (available upon request) for the rapid review was developed prior to the conduction of the review. However, this was not registered on PROSPERO, as it is not yet possible to register rapid reviews on this website.

Although the methodology adhered to these guidelines, it is worth emphasizing that corruption is a complex topic to research due to imbalances in power. This can lead to actors being reluctant to report corruption cases, or cases not being judicially solved due to a weak rule of law (18). Therefore, while the conducted methodology did provide an indication of how corruption unfolded in the selected countries, the authors acknowledge that it did not guarantee that all corruption cases were identified.

This review included four countries, two from WE and two from CEE. These countries were selected based on their geographical location (Western vs. Central-Eastern Europe), the contrast in corruption indicators (control of corruption index and rule of law index), the availability of information in English or Polish, and finally the authors' ability to contact experienced health experts from these countries to help identify additional cases.

The inclusion criteria for identified studies were determined following the CoCoPop (Condition, Context, Population) approach (19). Although CoCoPop was created for observational studies evaluating the prevalence or incidence of a specific disease, the authors considered it appropriate for the research's aim. This was because it aligned with the aspects under investigation. The elements of this mnemonic are stated as follows:

- Co: Health corruption.
- Co: Covid-19 pandemic.
- Pop: Selected WE and CEE countries.

In accordance with CoCoPop, the included studies must have addressed health corruption (condition) in any form. The European Commission typology (Table 1) was used to classify the cases of corruption, as the European Commission serves as a supranational ruling authority for all included countries. This ensured a comprehensive and standardized approach. For the context element, studies or reports must have been performed in the context of the COVID-19 pandemic. They must also have reported health corruption in an activity related to either preparedness for the pandemic or the response to it. For the population element, eligible studies referred to the selected WE (the Netherlands and the United Kingdom) and CEE (Poland and Slovakia) countries. The timeframe was limited to the period between March 11th 2020, when the WHO declared COVID-19 a pandemic (21), and April 15th 2021, when the last literature search was conducted. Only studies published in English and Polish were considered.

The study types that were considered for inclusion included abstracts, observational studies (e.g., case studies and case reports),

TABLE 1 Types of health corruption included in the review.

Corrupt practice	Subtypes
Bribery in medical service delivery	• Access to healthcare • Preferential treatment • Better quality of healthcare • False sick leave statements
Procurement corruption	• Pre-bidding: corruptive needs assessment • Pre-bidding: circumvention of tender procedures • Pre-bidding: tailored tendering • Bidding: bribery and kickbacks during the bid evaluation • Bidding: favoritism • Bidding: collusion and/or market division in bidding • Post-bidding: false invoicing • Post-bidding: changing contract agreements
Improper marketing relations Improper marketing regulations	• Direct prescription influencing (quid-pro-quo deals) • Indirect prescription influencing (creation of loyalty) • Undue positive list promotion • Authorization of medicines and certification of medical devices
Misuse of (high-level) positions	• Revolving door corruption • Regulatory state capture • Trading in influence • Conflict of interest • Favoritism and nepotism
Undue reimbursement claims	• 'Upcoding' (reimbursement of maximum tariffs) • Reimbursement of unnecessary treatments • Reimbursement non-delivered treatments
Fraud and embezzlement of medicines and medical devices	• Sale of public or prepaid medicines for private gain • Sale of counterfeit medicines • Use of publicly owned or financed devices or facilities for private gain

Source: Study on Health Corruption by the European Commission (20).

and reports available in the gray literature (policy briefs, statements from governmental and non-governmental organizations, news, and media reports). Health and politics-related databases were also consulted. These included Scopus, MEDLINE *via* WoS, and IBSS (International Bibliography of the Social Sciences) *via* ProQuest. Additional sources of information were also used, such as Transparency International and its relevant national chapters, the European Commission for reports from the Netherlands, Poland and Slovakia, and Anti-Corruption United Kingdom for information from the United Kingdom. As health corruption usually becomes known through media scandals, a Google search was also performed and limited to the first 50 results. Furthermore, we used the Web of Science to examine the references and track the citations of all included studies. Finally, we contacted health system experts from the four countries to ask for cases not identified through the literature search.

The following groups of terms and synonyms were considered for the search strategy: Health corruption, bribery, extortion, fraud, nepotism, racketeering; COVID-19, SARS-CoV-2, coronavirus; and The Netherlands, Poland, Slovakia, the United Kingdom. The search strategies for MEDLINE, IBSS, and Scopus, are presented in the Supplementary Tables S1–S3. An advanced Google search was performed using the same terms. Google Chrome's Incognito Mode was used during the Google search to avoid saved cookies and browsing history.

Two authors collaborated in the selection of the studies. AGA conducted the search strategy and selected studies for potential inclusion. Titles, abstracts, and full-text articles were then screened and evaluated by two authors (AGA and ACP). Decision disagreements were solved on consensus or by a third author (IKW). A PRISMA flowchart of the studies was constructed to illustrate the study selection flow. Due to the nature of reporting corruption cases, the authors anticipated most cases would be identified in gray literature, for which standard risk of bias assessments (e.g., ROBINS-I, Newcastle-Ottawa) would not be appropriate. Therefore, the Authority, Accuracy, Coverage, Objectivity, Date, and Significance (AACODS) checklist was conducted by two authors (AGA and ACP). This served to conduct a critical evaluation and determine the risk of bias from the identified studies (22). A third author (IKW) was consulted when discrepancies occurred.

The extracted information included: 1) a description of the case, 2) the country where it occurred, 3) the classification of health corruption using the European Commission framework (Table 1), 4) the actor from the HCS who was involved in the case based on Figure 1, 5) the date when the case occurred, 6) the date the case was acknowledged or identified, and 7) the person/organization who acknowledged or made the case public. One author (AGA) extracted the information, which was later reviewed by ACP. Discrepancies in the data extraction were achieved through consensus. Data synthesis was performed using the Synthesis Without Meta-analysis (SWIM) guidelines (23). The narrative synthesis was performed around the following themes: the types of health

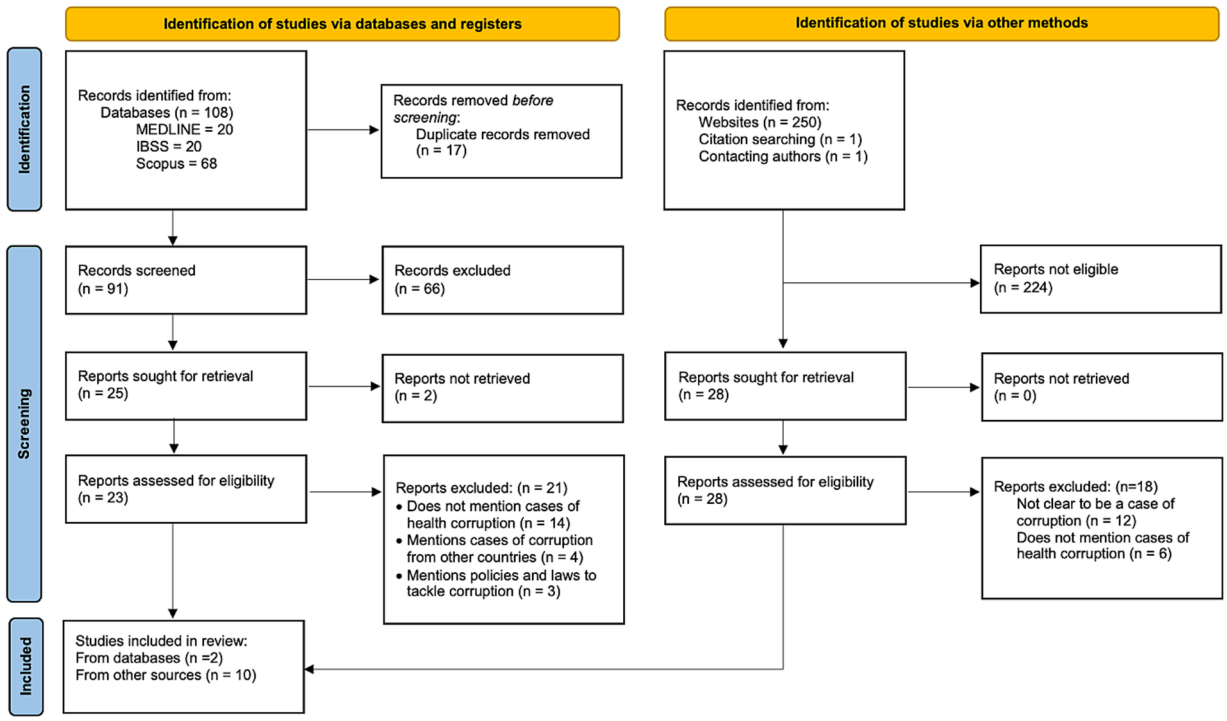

FIGURE 2
PRISMA flowchart of the included studies. Adapted from Page et al. (24).

corruption identified (10), the actor(s) involved in the identified cases, and the actor(s) that made the case public.

Results

Twelve studies were included in the final analysis. A critical appraisal of the included studies and the reference list of the excluded studies is presented in Supplementary Tables S4 and S5 of the supplementary material. Most were found through gray literature and the rest from opinion articles (Figure 2). From these, 13 cases were identified. The majority were from United Kingdom (6 cases - 46.2%), followed by Poland (4 cases - 30.8%), Netherlands (2 cases - 15.4%), and Slovakia (1 case - 7.7%). A summary of the description of the identified cases is shown in Table 2 and the results of the AACODS checklist for critical appraisal is presented in Table 3.

Types of health corruption identified

Three main types of health corruption were identified within the 13 cases: misuse of (high-level) positions (8 out of 13 cases); procurement corruption (7 out of 13 cases); and fraud and embezzlement of medicines and medical devices (5 out of 13 cases). In over half of the cases (53.8%), more than one type of health corruption was present. In WE countries, procurement corruption was present in 6 out of 8 cases. Fraud and the embezzlement of medicines/medical devices were reported in 3 out of 8 cases, while misuse of (high-level) positions also occurred in 3 out of 8 cases.

In the United Kingdom, for example, the National Audit Office reported that the UK government produced a "high-priority" list of suppliers with political connections (36). At the same time, cases of fraud were present as Public Health England paid for antibody kits that were later confirmed to be inaccurate (9). In the Netherlands, a case of procurement corruption was reported by Homolova and Lyndell (31), while Interpol detected an instance of fraud involving €1.5 million (30).

Contrastingly, CEE countries reported a higher number of cases in which the misuse of (high-level) positions was present (5 out of 5 cases). Fraud and the embezzlement of medicines / medical devices were present in 2 out of 5 cases, while procurement corruption was found in 1 out of 5 cases. Examples of the misuse of (high-level) positions occurred in Poland at the Military Institute of Hygiene and Epidemiology in Warsaw, as reported by the Central Anti-Corruption Bureau (28). In Slovakia, the Head of the Material Reserves acquired COVID-19 tests at 15 times the original price (29).

Actors of the HCS involved

Most of the actors involved in health corruption cases identified during the COVID-19 pandemic were authorities or organizations at central government level (9 out of 13 cases). This was followed by decentralized authorities (5 out of 13 cases), while only one supplier and one actor at the supranational level were involved among all 13 cases.

The actors at central government level were the most commonly involved in both WE (5 out of 8 cases) and CEE countries (5 out of 5

TABLE 2 Summary of findings from the included studies.

Author, year Country	Description of the case	Actor(s) of the HCS involved	Classification of health corruption	Date of occurrence and announcement	Responsible for the announcement
Kość (25) Poland	The public health minister spent 5 million złoty (€1.1 million) on 120,000 FFP-2 type face masks and 20,000 surgical masks that were later found not to meet safety standards. The contract originated from a ski instructor who knew the actor's brother, a health care businessman.	• Central government, Health Ministry	• Fraud and embezzlement of medicines and medical devices. • Misuse of (high-level) positions.	Occurrence: Not clear Announcement: Not clear	Newspaper, Wyborcza
Koper (26) Poland	A former deputy health ministry agreed to pay €35 million to an arms dealer for ventilators without experience in health devices. Normally, the authorities hold public tenders for such large deals. In March, Poland's parliament passed a law saying that tenders are unnecessary in COVID times.	• Central government, Deputy Health Ministry	• Fraud and embezzlement of medicines and medical devices. • Misuse of (high-level) positions.	Occurrence: April 14th, 2020 Announcement: December 17th, 2020	Newspaper, Reuter
ENCA (27) Poland	Politicians, including members of the ruling Law and Justice (PiS), a Member of the European Parliament, and the former Prime Minister, received their COVID-19 vaccine ahead of their turn.	• Supranational organization, Member of European Parliament. • Central government, members of the ruling party.	• Misuse of (high-level) positions.	Occurrence: December 30th, 2020 Announcement: January 4th, 2021	Newspaper, ENCA
Central Anti-corruption Bureau (28) Poland	Between March and June 2020, the Director of the Military Institute of Hygiene and Epidemiology in Warsaw abused his powers by appropriating property belonging to the Institute. This property included protective equipment, disinfectant fluids, and tests for COVID-19. The investigation indicates that those items were then transferred to unauthorized persons.	• Central government institution, Director of the Military Institute of Hygiene and Epidemiology in Warsaw.	• Misuse of (high-level) positions	Occurrence: Not clear Announcement: Not clear	Central Anti-corruption Bureau
Kern (29) Slovakia	The Head of the Material Reserves office purchased 200,000-speed tests of lower quality for the novel coronavirus for €6 million, 15 times more than what China paid. Also, he received a €200,000 transfer to his account, suspected to be from bribery.	• Central government, Head of the Material Reserves	• Procurement corruption • Misuse of (high-level) positions	Occurrence: April 21st, 2020 Announcement: April 21st, 2020	Police from Slovakia
Interpol (30) Netherlands	Interpol detected a scheme to defraud German authorities in transactions to purchase face masks. A Dutch supplier requested €1.5 million in advance, claiming that the funds were not transferred, and an additional €880,000 were demanded to secure the shipment.	• Supplier, a Dutch company.	• Fraud and embezzlement of medicines and medical devices	Occurrence: March 15th, 2020 Announcement: April 15th, 2020	Interpol
Homolova and Lyndel (31) Netherlands	Information about the contracts awarded for purchasing goods related to the COVID-19 pandemic has not been made public. This includes the names of the companies awarded the contracts and the amount received.	• Central and/or decentralized government.	• Procurement corruption	Occurrence: Not clear Announcement: October 21st, 2020	Organized Crime and Corruption Reporting Project

(Continued)

TABLE 2 (Continued)

Author, year Country	Description of the case	Actor(s) of the HCS involved	Classification of health corruption	Date of occurrence and announcement	Responsible for the announcement
Goodrich (32) United Kingdom	A total of 24 PPE contracts, accounting for £1.6 billion, were awarded to those with known political connections to the Conservative Party, and three contracts worth £536 million went to politically connected companies for coronavirus testing services.	• Central government members of the ruling party.	• Misuse of (high-level) positions. • Procurement corruption.	Occurrence: February 2020 to December 2020 Announcement: April 22nd, 2021	Transparency International
National Audit Office (33) United Kingdom	"PPE suppliers with political connections were directed to a "high-priority" channel for United Kingdom government contracts where bids were ten times more likely to be successful. Almost 500 suppliers with links to politicians or senior officials were referred to the channel, where their pitches for contracts were automatically treated as credible by government officials charged with procuring PPE.	• Central government, Department of Health and Social Care, Department of Education, and Cabinet Office	• Misuse of (high-level) positions. • Procurement corruption.	Occurrence: From March 2020 to July 2020 Announcement: November 26th, 2020	National Audit Office
BBC (34) United Kingdom	A court ruled that the Secretary of State for Health and Social Care acted unlawfully by not revealing the information about the contracts it had signed during the COVID-19 pandemic.	• Central government, Secretary of State for Health and Social Care	• Procurement corruption.	Occurrence: From March 2020 to December 2020 Announcement: February 19th, 2021	High court
Kohler and Wright (9), United Kingdom	A senior NHS official in London working at the capital's Covid-19 Nightingale hospital launched a business in April 2020 to trade PPE. The official was suspected to be part of procurement for PPE in the NHS.	• Decentralized authority, NHS official	• Misuse of (high-level) positions. • Procurement corruption.	Occurrence: April 15th, 2020 Announcement: May 1st, 2020	News, The Guardian
Kohler and Wright (9) United Kingdom	Public Health England paid for antibody kits for COVID-19, but they later proved inaccurate.	• Central government, Public Health England	• Fraud and embezzlement of medicines and medical devices.	Occurrence: April 1st, 2020 Announcement: April 11th, 2020	Greg Clark, Chair of the Commons science and technology committee
Armstrong (35) United Kingdom	The United Kingdom government awarded a contract of £75 m for one million antibody tests to a business consortium. A public tender was not carried out, and the accuracy of the tests was not evaluated.	• Decentralized authorities, United Kingdom Rapid Test Consortium	• Fraud and embezzlement of medicines and medical devices. • Procurement corruption.	Occurrence: October 6th, 2020 Announcement: November 12th, 2020	Good Law Project

PPE, Personal protective equipment.

cases). Decentralized authorities were involved in one case in WE (9). An authority from a supranational organization was involved in one case in Poland (27), while a supplier was responsible for a case of health corruption in the Netherlands (37).

Authority or organization that made the case public

From the 13 cases, four were reported by the news, three by the rule of law authorities, three by non-governmental anti-corruption organizations, two by the police, and one by a government anti-corruption watchdog. The organization that made the cases public differed between WE and CEE countries. For instance, in the United Kingdom and the Netherlands, three cases were reported by an anti-corruption NGO (Good Law Project, Transparency International, and the Organized Crime and Corruption Reporting Project), three by the rule of law (National Audit Office, Chair of the Commons, and High Court), one by the media, and one by the Interpol. In CEE, the authorities reporting cases of corruption during the COVID-19 pandemic were mainly the media (3 out of 5 cases), one by the police, and only one by an anti-corruption watchdog (28).

TABLE 3 AACODS critical appraisal based.

	Authority	Accuracy	Coverage	Objectivity	Date	Significance
Kość (25)	?	Y	Y	Y	Y	Y
Koper (26)	Y	Y	Y	Y	Y	Y
ENCA (27)	N	?	Y	Y	Y	Y
Central Anti-corruption Bureau (28)	Y	Y	Y	Y	Y	Y
Interpol (30)	Y	Y	Y	Y	Y	Y
Homolova and Lyndel (31)	Y	N	?	?	?	Y
Kern (29)	Y	Y	Y	Y	Y	Y
Goodrich (32)	Y	Y	Y	Y	Y	Y
National Audit Office (33)	Y	Y	Y	Y	Y	Y
BBC (34)	Y	Y	Y	Y	Y	Y
Kohler and Wright (9)	Y	Y	Y	Y	Y	Y
Kohler and Wright (9)	Y	Y	Y	Y	Y	Y
Armstrong (35)	Y	Y	Y	Y	Y	Y

Discussion

Our research presented the results of a rapid review of the literature used to identify, describe, and categorize health corruption cases in WE and CEE countries during the COVID-19 pandemic. The findings showed that corruption was present in each of the studied countries. Evaluations of corruption during the pandemic are still emerging among European countries, with the number of reports expected to increase as data on procurement and resource allocation is analyzed by researchers, media, and watchdogs. Recent literature in the field has explored the impacts of corruption on public trust and mortality due to COVID-19 (37, 38).

As of the writing of this article, Europe faces fresh shocks in the form of the war in Ukraine, the refugee flow, and the need to create decent and equitable conditions of stay for the individuals concerned. As demonstrated during the COVID-19 pandemic, each crisis increases the potential for corrupt activities. Studies in this area are extremely important, particularly as cases of corruption related to these crises continue to be brought to light (39, 40).

Our review found the misuse of (high-level) positions to be the most prevalent type of health corruption in CEE countries during the COVID-19 pandemic. Procurement corruption was the main type encountered in WE countries. This was also the case prior to the pandemic (41). The involvement of actors from high-level positions in CEE countries is well documented in the literature. It is explained by the small, tight-knit friendship networks formed in the political sphere during the communist and post-communist eras, which facilitated opportunities to engage in these practices without being penalized (42). Meanwhile, better enforcement of the rule of law might explain the higher prevalence of procurement corruption found in WE countries (43).

The OECD has identified public procurement as one of the most vulnerable governmental activities. This is due to the high volume of transactions, financial interests, the complexity of the process, close interaction between public and private sector officials, and the multiple stakeholders involved (44). In the context of the COVID-19 pandemic, procurement was one of the first actions taken by the government to obtain the medical equipment required to address the pandemic (i.e., personal protective equipment (PPE), ventilators, etc.). Not surprisingly, most health corruption cases occurred when the government acquired these goods. In the United Kingdom alone, Transparency International identified 65 "questionable contracts" geared toward acquiring PPE, for which the government paid £2.9 billion (11). In addition to the inherent complexity of public procurement, governments approved laws that reduced the transparency of purchases during the pandemic. For instance, in the United Kingdom, the government introduced a "high-priority" channel for government contracts (9). In Poland, policies were implemented to give impunity for decisions taken to protect the population during the COVID-19 pandemic, even if these actions were not legal (45).

It is easy to understand the need to streamline the process for procuring medical goods during the COVID-19 pandemic, as it helped to ensure a rapid response to the crisis. However, corruption's long-term negative impact on the health sector might eventually outweigh the benefits, resulting in a lack of trust from the public sector. The consequences of distrust are directly associated with the public's behavior in response to a crisis. This was shown by Han et al., who reported that higher trust in government was reflected in the public's compliance with COVID-19 protection measures (handwashing, avoiding crowded spaces, self-quarantine, etc.). This also shows the fundamental importance of strong levels of trust before the onset of a crisis. As evidenced during the pandemic, the populations of countries with higher trust at baseline were far more likely to follow the recommended safety measures (46).

In all four countries, actors from the central government were the main ones involved in the identified cases. However, the mechanisms to report these cases differed among WE and CEE countries. Anti-corruption organizations reported most cases in WE countries, while media scandals were most the most frequent method of unearthing corruption in CEE countries. The centralized use of resources might explain the central government's involvement in most cases. This is shown by the recorded subnational government spending in each country: less than 10% in the Netherlands, Slovakia and the United Kingdom, and less than 20% in Poland (47).

The close networks of friends among the political authorities might hinder the reporting of health corruption in CEE, thus making

it necessary to establish effective whistleblowing mechanisms (38). Efforts to improve whistleblowing and protect those reporting corruption have been made at the supranational level, as evidenced by the European Parliament's Directive on protecting persons who report breaches of Union Law. This directive was expected to be adopted by all member states before the end of 2021. However, at the time of writing (August 2022), only 11 out of the 27 member states have adopted this law. The Netherlands, Poland and Slovakia are among those delaying its implementation (48).

Implications for policy and research

The implications for policy include the need to create appropriate anti-corruption mechanisms, as well as to implement anticipatory mechanisms for the rapid identification of cases of corruption. These anti-corruption mechanisms should prioritize safe whistleblowing to ensure that health system actors are protected and empowered to report corrupt practices. It is also important to enhance transparency and accountability for public procurement. A solution that has been successfully implemented in other European countries is open contracting for health (OC4H). OC4H is grounded in the wisdom that prevention is better than cure, and that the beneficiaries of the procurement (citizens, populations, patients) are in a better position to monitor the process (49). Lastly, anticipatory governance also plays a vital role in the prevention of corruption. Anticipatory governance aims to foresee and prevent the (unintended) negative consequences of policies using a whole-of-system and whole-of-government approach (50).

One implication for future research is found in the role our study played in identifying and categorizing cases of corruption, while also highlighting the differences between Western and Central-Eastern European countries. Our study brought into focus the contrast between both regions, demonstrating the need to individually tailor the ways we study and tackle corruption to best suit each country. Moreover, as cases of corruption continue to be reported, comprehensive reviews involving greater numbers of countries are needed. Reviews could be complemented by interviews with stakeholders, helping to identify further challenges and solutions regarding the uncovering and reporting of corruption.

Limitations

This study was constrained by notable limitations and the findings should be interpreted with caution due to the nature of the topic. We followed a systematic approach using standardized methods to explore a complex subject. However, the political implications of corruption and its sensitivity pose significant limitations to this study and following a systematic approach is not sufficient to capture all the corruption cases during the studied period. Thus, the study's results should be taken as additional research that contributes with evidence to the field of healthcare corruption rather than a solid conclusion on the topic. As mentioned previously, the nature of corruption makes it challenging to identify and report in the first place. The number of cases is limited due to their political implications and the fear of consequences and retaliation to report the cases. This limitation is widely recognized in the field of corruption in the healthcare sector (51). Publication bias places an important role in this matter. Although in health research publication bias refers to the selective publication of findings (52), in the study of health corruption this is related to the overall lack of reporting due to the safety implications mentioned before. Moreover, the review was conducted during the early stages of the pandemic, which might provide a limited picture of the overall number of cases. This is because, as stated earlier, new cases are expected to be reported as governments' decisions are scrutinized more closely.

Additionally, all information was limited to the English and Polish languages. Although we tried to overcome this limitation by contacting health system experts with experience in corruption research from each studied country to help detect additional cases, there is no guarantee that the experts were aware of all the cases. Therefore, the findings from non-English and non-Polish speaking languages should be interpreted cautiously as we might have missed other corruption cases. Finally, we did not explore the juridical verdict of the identified cases, as these processes are usually lengthy, and their outcomes depend mainly on the country's rule of law. Instead, we limited our analysis to identifying, describing, and categorizing corruption cases in the studied countries.

Conclusion

Cases of corruption in the health care sector were present in all four studied countries. However, the types of corruption differed in each country, with a higher prevalence of procurement corruption in WE countries and misuse of high-level positions among CEE. While a rapid response is necessary to deal with a shock like the COVID-19 pandemic, countries' efforts should focus on increasing the health systems' resilience by ensuring adequate resources and tackling corruption. As other crises emerge across Europe, corruption threatens countries' success in implementing effective responses. Thus, further research in preventing and tackling corruption is a vital and necessary undertaking despite the inherent limitations of conducting health corruption research.

Author contributions

AG-A: Conceptualization, Formal Analysis, Investigation, Methodology, Validation, Writing – original draft. AC-P: Data curation, Formal Analysis, Writing – review & editing. IK-B: Investigation, Supervision, Validation, Writing – review & editing.

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Supplementary material

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Considering patient perspectives in economic evaluations of health interventions

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Current guidelines for evaluating the cost-effectiveness of health interventions commonly recommend the use of a payer and/or a societal perspective. This raises the concern that the resulting reimbursement decision may overlook the full spectrum of impacts and equity considerations. In this paper, we argue that a potential solution is to supplement a societal- or payer-perspective economic evaluation with an additional evaluation accounting for exclusively the patient perspective. We present five categories of health interventions for which a patient-perspective analysis may be informative including those (1) that cross the definitional boundary between drugs and non-drug technologies; (2) affect patient adherence to protocol; (3) represent revolutionary treatments for genetic disorders; (4) with an incremental cost-effectiveness ratio involving slightly less effective, but substantially less costly, than the current standard; and (5) have been previously approved for funding but now being targeted for potential delisting or disinvestment. Real-world examples are discussed in detail. Lived experience individuals were invited to provide vignettes. Discussions are provided regarding how to incorporate patient inputs to improve patient-centered decision-making.

KEYWORDS

health economics, cost-effectiveness analysis, economic evaluations, patient-centered care, patient preference

1. Introduction

Economic evaluations, as reviewed by Turner et al. in an earlier issue of *Frontiers in Public Health*, refer to a type of analysis that simultaneously assesses the costs and effects of alternative interventions to ensure value for resources expended from various perspectives (1). Although there have been attempts to address equity considerations in cost-effectiveness analyses (CEAs) through the development of distributional and extended cost-effectiveness methods, they stated that "this is still an area that needs attention regarding practical implementation with regards to informing resource allocation decisions in global health" (1). This shortfall emanates, in part, from inadequate analysis of the burdens placed on individuals and families from the introduction of new health interventions.

Commonly, only the societal and/or the payer perspective are recommended by health technology assessment (HTA) agencies. At the national level, the latter is endorsed by countries including Australia, the UK and Canada (2). Other countries (e.g., Thailand) have recommended a societal perspective while at least one—Norway—has limited it to exclude

productivity gains and losses (2). The US has joined a small number of countries (e.g., Italy) recommending both perspectives as reference cases (3).

A pressing issue with such perspectives is that they may insufficiently highlight impacts on lived experience individuals, including patients and families/caregivers (henceforth "patients" for simplicity) (4). Although the societal perspective can capture some patient-borne financial impositions, they are aggregated with other costs, raising the concern that a favorable societal-perspective profile may not signal whether for patients an intervention is truly affordable, adherable or otherwise impactful (5). The affordability concern is accentuated when assessing innovative health interventions that straddle the definition of prescription drugs and non-drug technologies because the level of third-party coverage is *a-priori* unclear; therefore, patients may face substantial increases in out-of-pocket costs if an intervention is assigned to a category with lower levels of coverage than its predecessors (5). These interventions may also impose negative impacts on patient lives that would remain "hidden" within broader perspectives.

Herein, we argue that a potential solution—of particular interest to public health professionals—is to supplement a societal- or payer-perspective economic evaluation with an exclusive evaluation from the perspective of patients. While guidelines for incorporating patient inputs in HTA exist (6–8), a formal framework for conducting patient-perspective economic evaluations has never been formulated to our knowledge. As such, CEAs conducted with the patient perspective often vary in their coverage and definition of patient-borne costs (4). In this Perspective paper, we aim to provide a comprehensive discussion on the unique insights a patient-perspective economic evaluation could yield to aid reimbursement decision-making in healthcare.

2. Procedures

In this 2-part Perspective paper, we followed the Patient Preferences in Benefit–Risk Assessments during the Drug Life Cycle project and a subsequent focus group discussion with HTA representatives from Canada, Belgium and Germany (9, 10). In the first part of this paper, we formulated five categories of health interventions that could be used to flag when a supplementary analysis might be warranted through extensive discussion within our team. The issues covered include household-level affordability; adherence; unintended side effects; and burdens on daily living. A separate category was created for gene therapy as it has been regarded as a health technology particularly sensitive to the patient perspective (9, 10). Real-world examples are discussed.

To provide real-world perspectives on this topic, during December 2020, we invited lived experience individuals from the Patient and Family Advisors Network, a virtual network that comprises individuals covered under Ontario's publicly funded healthcare insurance system who volunteer to serve as policy advisors for Ontario Health, to review the draft of this paper. Using a convenience sampling methodology, one researcher (DW) sent out email invitations where five individuals ultimately responded and agreed to review this paper. We present the comments from the 5 reviewers in Box 1. We conclude this paper with further discussions regarding when to conduct a patient-perspective analysis alongside some cautionary notes. Currencies were adjusted to 2020 using the annual Consumer Price Index and then converted to US dollars using purchasing power parities (11).

3. Types of health interventions that may warrant a patient-perspective analysis

3.1. Interventions that cross the definitional boundary of drugs and non-drug technologies or replace prescription drugs

The level of third-party coverage for drugs and non-drug technologies varies, most likely due to differing societal preferences for coverage and decision-making mechanisms. As one of the first countries to institutionalize HTA, Canada has operated a centralized evaluative process for recommending funding for brand-name drugs since 2003 through the Canadian Agency for Drugs and Technologies in Health's Common Drug Review. Funding recommendations regarding non-drug technologies are made solely at the provincial level with notable variability in processes and decisions (12). This fragmentation creates issues for an emerging class of products that can neither be classified as a drug or non-drug technology. Patients who seek access to these products face financial obstacles as third-party coverage may have varying criteria for coverage; one of our reviewers shared this sentiment (Box 1). At the top of the scale, prescription drugs are generally covered within a single program budget with higher levels of coverage than within program budgets that cover medical devices (13). Thus, we believe funding decisions regarding these novel products might benefit from a separate patient-perspective analysis as it may highlight the potentially large increase in out-of-pocket costs. We present the examples of EndeavorRx, a game-based digital therapeutic devise approved in the US, and Abilify MyCite, a prescription drug with a digital ingestion tracking system, under this category.

The US Food and Drug Administration (FDA) has recently approved interventions that either replace prescription drugs or represent a hybrid of a drug and a medical device. An example of the first, EndeavorRx, was approved in 2020 as the first game-based digital therapeutic device prescribed for children with attention deficit hyperactivity disorder. This mobile device-based game involves navigating through a course with obstacles that is designed to improve attention function. EndeavorRx obtained FDA clearance through the *De Novo* Classification Pathway due to the lack of a predicate device on the market. To date, neither the FDA nor the manufacturer have released information on pricing and insurance coverage; hence, it is unknown what share of the cost families will face for this first-of-its-kind therapeutic device.

In 2017, Abilify MyCite was granted US market entry as the first prescription drug with a digital ingestion tracking system. Intended to treat adults with schizophrenia, bipolar disorder and depression, this combination product consists of Abilify (aripiprazole) tablets with an ingestible sensor that sends signal to a wearable patch communicating with a smartphone. Priced at 1,650 USD per month, Abilify MyCite is 80-times more costly than its drug-only counterpart and is covered in the US only for the most economically challenged (Medicaid beneficiaries). The cost-effectiveness of Abilify MyCite to the payer/society has yet to be established, but with its high

BOX 1 Comments received from lived experience individuals.

Interventions	Comments
Interventions that cross the definitional boundary of drugs and non-drug technologies or replace prescription drugs	*"We too are often bewildered on how to engage these new "cross-program" interventions."*
Interventions that significantly affect patient adherence to protocol by having unintended side effects	*"As a caregiver for a child with severe scoliosis, I endured a daily struggle to try to put him in a plastic back brace leaving me heartbroken as I just could no longer force him to wear it. I was also consumed with guilt because without the brace the alternative would be spinal surgery with all of the added risks involved."* *"Adherence is ultimately in the sphere of influence controlled by the patient. It therefore runs completely counter-intuitive for patients not to be engaged in interventions that impact this critical nexus."*
Interventions that represent revolutionary treatments or even cures for genetic disorders	*"Is the revolutionary intervention truly a cure or does it just hide the genetic disorder or just make it livable?"*
Interventions previously approved for coverage but now targeted for potential delisting or disinvestment	*"This is an area that I get most comments from other caregivers. They do not understand how their child, youth or adult can be deprived of a treatment that was working."*

out-of-pocket costs, it is unlikely to be affordable to many households without substantial third-party coverage.

3.2. Interventions that significantly affect patient adherence to protocol by having unintended side effects

A health intervention that is likely to impact adherence and priced substantially more than standard treatments warrants scrutiny within the entire HTA process. A common method to account for non-adherence in CEAs is through a small decrement in efficacy to produce a *de facto* measure of effectiveness; however, the decrement is commonly too small to result in any meaningful change in an incremental cost-effectiveness ratio (ICER) that would impact HTA recommendations (14). One of our reviewers, who are actively caring for a child wearing a medical device, expressed frustration in witnessing the side effects of the device and the lack of a more adherable and comfortable alternative (Box 1). Meanwhile, adherence-enhancing interventions usually demonstrate superior cost-effectiveness—if not cost-saving—from the societal or payer perspective (15). However, negative impacts on patient lives may remain essentially hidden within broader perspectives as there may be substantial benefits within these perspectives that could offset these negative impacts to patients; therefore, patient-perspective evaluations are needed to reveal potentially important patient-level effects; one of our reviewers concurred (Box 1).

One example of such an intervention—FreeStyle Libre—is a wearable flash glucose monitoring system for people with diabetes as an alternative to finger-prick tests. The system comprises a disposable subdural sensor and a device that receives and stores data. In 2020, the Ontario Health Technology Advisory Committee (OHTAC) and the HTA Advisory Board of Quebec have both recommended funding FreeStyle Libre, despite analyses showing that it may not be cost-effective to payers compared to finger-prick tests (16). A recommendation to fund the intervention was partially driven by feedback from patients and caregivers who reported greater ease of use permitting them greater control over their lives. This helped to overcome uncertainty over its cost-effectiveness to improve blood glucose stability.

While Abilify MyCite has already been mentioned, there is a general class of drug-device hybrid products called digital pills emerging that combine prescription drugs with an ingestible sensor, a wearable patch and a mobile application (17). By continuously tracking patients' medication-taking behaviours, digital pills aim to improve patient adherence, help forge self-care routines and enhance patient-physician relationships. However, patients report fatigue and disruption of daily routines by going through extensive training to correctly operate the wearable patch and to successfully pair it with a smartphone. This imposition on patient lives may be greater than the burdens of manually remembering to take medications thereby defeating the marginal benefit associated with using digital pills (18). Another issue is related to privacy, that is, who would be authorized to access patient data. At the extreme, these concerns may be disruptive leading to a refusal to take the medication. Furthermore, there is the danger of device-associated emergent adverse events that have implications for both patient well-being and adherence to protocol (17). These uncertainties raise questions regarding willingness-to-pay out-of-pocket for a product carrying such risks for improvements in adherence. A formal patient-perspective analysis may be able to provide answers by soliciting inputs that highlight these uncertainties.

Nirmatrelvir with ritonavir, sold under the brand name Paxlovid, is another example of interventions that fall under this category. In December 2021, the US FDA approved the use of Paxlovid for treating mild-to-moderate COVID-19 under an emergency authorization. Soon after its approval, reports of side effects started to emerge, most notably regarding the bitter metallic taste Paxlovid sometimes left in mouth ("Paxlovid mouth"). Recent studies of "Paxlovid rebound," which refers to an asymptomatic or symptomatic resurgence of COVID-19 after finishing the full 5-day course of Paxlovid, suggest this may be due to patients skipping a dose to avoid the unwanted aftertaste of Paxlovid (19, 20). This conjecture requires more research to confirm.

3.3. Interventions that represent revolutionary treatments or even cures for genetic disorders

Recent breakthroughs have enabled new treatments for genetic disorders that previously were considered untreatable. In clinical trials, these treatments demonstrate high incremental effectiveness or even a cure. While revolutionary, they may place added burdens on patients' lives or produce inadequately measured risks. Indeed, one of our reviewers raised concern on the real-world outcome of these treatments, and specifically, if a cure was truly attainable (Box 1). In this case, we suggest that a separate patient-perspective analysis that elicits inputs from patients and their families might provide important insights to support informed decision-making. We talk about treatments for spinal muscular atrophy as an example.

In 2017, Spinraza (nusinersen) was approved by the US FDA and the European Medicines Agency for the treatment of spinal muscular atrophy, a group of rare, genetic neuromuscular disorders that leads to severe muscle weakness and progressive loss of motor function. In 2020, The Japanese Ministry of Health, Labour and Welfare approved Zolgensma (Onasemnogene abeparvovec), a single-dose intravenous gene replacement therapy that replaces Spinraza's four loading doses. Compared to Spinraza, Zolgensma is less costly (2 million USD vs. 2.2–10.6 million USD for Spinraza over a lifetime) and is potentially superior to Spinraza by reducing treatment complexity (21). However, patient representatives have voiced concerns on the durability of the long-term benefits of Zolgensma and on the uncertainty of treatment pathways if gene expression diminishes over time (21). Furthermore, to help better understand the particulars of treatment protocols, patients may value genetic counseling, but the availability of such counseling service is often limited in real-world clinical settings—a usually unmeasured shortcoming. None of these effects are captured within current analyses.

3.4. Interventions with an expected ICER in the southwest quadrant of the cost-effectiveness plane from a payer or societal perspective

Funding decisions are often difficult for interventions that are less costly but slightly less effective from payer/societal perspectives than currently utilized therapies. Debates have been on the extent to which the loss of a quality-adjusted life year (QALY) differs from the value of acquiring a QALY. Review studies found Willingness-To-Accept/Willingness-To-Pay ratios among health interventions to range from 1.9 to 6.4 (22). These observations point to a kink in consumer threshold values where patients are generally more reluctant to lose than they are willing to gain. Currently, there is a lack of consensus on how to address this potential asymmetry (23). Hence, we argue that when confronted with this situation, a patient-perspective analysis in which patient-level costs are weighted more heavily might help guide funding decisions.

In 2002, the National Institute for Health and Care Excellence (NICE) recommended vinorelbine as one option for second-line treatment for advanced breast cancer (24). The only UK-based CEA at that time concluded vinorelbine to be slightly inferior to taxanes in terms of quality of life improvement, while being markedly cheaper. The same study found the ICER of vinorelbine to be 37,277 USD/QALY (14,500 pounds/QALY in 1998 values) and 5,116 USD/QALY (1,990 pounds/QALY in 1998 values)—in the southwest quadrant—when compared to docetaxel and paclitaxel, respectively. Taking into account the patient perspective, NICE recommended to keep funding vinorelbine due to its safety and tolerability among patients distinguishing vinorelbine as a more patient-friendly option than taxanes.

An internet-mediated cognitive behavioral therapy for treating mild to moderate depression is a more recent example (25). Trial results suggest that this Swedish-recommended intervention produced 0.05 fewer QALYs over 12 months while saving 326 USD (2,664 SEK in 2013 values) for the society compared to usual care (primary care physician visits, nurse visits, antidepressants, face-to-face psychotherapy and/or sick leave). The study concluded that this therapy is at least as cost-effective as usual care and that the choice of treatments ultimately relied on patient preferences regarding ease-of-use, availability and willingness to wait for services.

3.5. Interventions previously approved for coverage but now targeted for potential delisting or disinvestment

Third-party payers sometimes must make decisions to delist a previously reimbursed intervention. This may have profound and sometimes unintended consequences to patients. According to one of our reviewers, caregivers are usually baffled by the delisting of a health intervention that has been working for the patient (Box 1). It is imperative that, prior to delisting, the perspective of patients' needs to be considered to avoid harm to subsets of patients. Furthermore, a grace period during which patients can be phased out of these interventions and transition into a new protocol should be clearly defined and structured. We argue that a patient-perspective analysis may help clarify the relative costs and benefits that will befall patients.

In 2017, the OHTAC recommended discontinuing public funding for external cardiac loop recorders (ELR) for diagnosing cardiac arrhythmia if the device relied solely on patient-initiated recordings. In consideration of the recommendation, the Ontario Ministry of Health now funds ELR only if it is operated by a cardiologist and funding also continues for long-term continuous ambulatory electrocardiogram monitors (ECGm), a more advanced alternative (26). These decisions were made on the basis that, despite a small annual increase in provincial expenditures due to increased diffusion of ECGm, these expenses were justified given incremental improvements in patient experiences. However, cardiologists voiced concerns regarding the disinvestment decision as ELR and ECGm had been traditionally used to diagnose different patient populations—one that is able to self-monitor and the other monitored by a cardiologist, respectively. Hence, a phasing out of reimbursement for ELR reduces patient access to a proper diagnosis as a limited supply of cardiologists must now supervise ELR testing (27). This issue cannot be overlooked because adoption and disinvestment policies need solid implementation plans, including potential grace periods, to manage unintended consequences in both cases.

These unintended effects of disinvestment on patients and their caregivers should be better anticipated. For example, expectorants and mucolytics, that make coughing up mucus easier and less irritating,

were dropped in France from the publicly funded formulary in 2006 as a physician's prescription was no longer needed. The resulting over-the-counter market for these products saw price increases of up to 200%, the full cost of which was borne by patients leading to affordability issues for those with lower incomes (28). A second example involved the delisting of phlebotonics for chronic venous diseases in Italy that was followed by an increase in hospitalizations for venous insufficiency (29). Perhaps, better consultations with experts could assist implementation to either soften or prevent the adverse effects noted.

4. Discussion

Existing health economic evaluation studies using the patient's perspective rarely provide a clear rationale on the conduct of a focused patient-centred examination (4). As such, the preceding has been a listing of situations in which patient-level assessment of both affordability and level of imposition on patient lives is warranted alongside a societal/payer perspective analysis to partially address equity concerns. In terms of affordability, many jurisdictions provide full coverage for those under the poverty line. For others, a limit is set on out-of-pocket costs between 1% to 7.5% of income in order to qualify for either tax deductions or credits, and/or government programs that provide catastrophic coverage (30, 31). Thus, affordability varies by level of income under the current patchwork system (32). A marginal increase of at least 1% of income over standard praxis might be worthy of notice for any subset of patients based on the base threshold set for tax deductibility of medical expenses in Germany, the lowest amongst western countries (31). No similar threshold for imposition on patient lives neither exists nor is recommended other than to suggest assessing the impacts individually for each intervention.

Though perhaps not exhaustive, the list of situations provided represent a first attempt to elucidate a shortcoming in current economic evaluation guidelines. We have shown that there is the potential for substantial incongruencies in findings between narrower economic analyses from the perspective of patients than broader analyses. These differences in findings may suggest that net effects in these broader perspectives tend to hide details that may be important from the point-of-view of decision-making bodies. By weighing all costs equally, these broader analyses may be underestimating some cost components that these bodies may wish to weigh more heavily prior to concluding about whether and how to move forward. We believe HTA agencies need to actively incorporate patient inputs throughout the entire HTA process, but until we reach expert consensus on how to quantitatively account for these data in cost-effectiveness analyses, more feasible options include a qualitative literature review about patient preferences and/or direct engagement activities (such as a focus group discussion) with these individuals (33). Caution must be taken, however, to not double-count time/adherence costs that may already be captured within QALY decrements and therefore should not be added to financial costs. At this juncture, more work is needed to explicate the various situations in which a patient-perspective economic evaluation is not only warranted but potentially recommended.

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

RF, VN, ML, DW, EY, and EN: conceptualization, investigation, and writing—review and editing. RF, VN, and EN: methodology. RF: writing—original draft preparation. EN: supervision. All authors contributed to the article and approved the submitted version.

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Conflict of interest

VN was employed by Roche Diagnostics.

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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Optimizing internal control in public hospital supply chain: a game theory-based approach

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Objective: Our study aims to enhance the precision of internal control construction within public hospital supply chains and minimize the subjective bias influence. We have integrated the game theory combination weighting method into the design of internal control paths and based on this, developed a series of pioneering solutions. This innovative approach is anticipated to heighten the effectiveness and scientific rigor of the internal control design scheme within the supply chain.

Method: Firstly, we utilized literature review and expert interviews to delve into the key factors of public hospital supply chain internal control, forming an index system for public hospital supply chain internal control that aligns with current informatization requirements. Subsequently, we incorporated the Game Theory Combination Weighting Method into this study. By means of the Analytic Hierarchy Process and the Entropy Weighting Method we determined the subjective and objective weights of each index and obtained their comprehensive weights through the Game Theory Combination Weighting Method. Then, based on the analysis results, we designed a series of internal control construction schemes and implemented these schemes at Weifang Maternal and Child Health Hospital between 2019 and 2023. Finally, using the Fuzzy Comprehensive Evaluation Method to assess and compare the actual effects before and after the implementation of the schemes, thereby validating the effectiveness of the Game Theory Combination Weighting Method in the design of the internal control path of public hospital supply chains.

Results: The fuzzy comprehensive evaluation results for the years 2019 and 2023 demonstrated that after implementing our design schemes using the Game Theory Combination Weighting Method, the hospital's satisfaction in aspects such as plan-side control, purchase-side control, asset-side control, expenditure business control, and contract management control has significantly improved.

Conclusion: Our research indicates that the Game Theory Combination Weighting Method is applicable to the path design of internal control links in public hospital supply chains. This method has effectively enhanced the targeted improvement of weak links within the construction of internal controls in the supply chain of public hospitals and is of great significance for improving the scientific nature of supply chain internal control management.

KEYWORDS

internal control, supply chain, combination weighting method of game theory, entropy weighting method, analytic hierarchy process

1. Introduction

The more widely used definition of internal control is proposed by the National Anti-fraudulent Financial Reporting Committee (Committee of Sponsoring Organizations of the Treadway Commission, COSO Committee) in "Internal Control: An Integrated Framework" (1), that is, internal control is the process of providing reasonable assurance for the achievement of objectives such as operating results, authenticity of financial reports and compliance with applicable laws. The supply chain is an integrated manufacturing process wherein raw materials are converted into final products, then delivered to customers (2, 3). The effectiveness and efficiency of the supply chain are largely contingent upon its internal control mechanisms. Internal controls not only ensure the smooth operation of each link in the supply chain but also offer risk management and decision-making support for supply chain management, thereby guaranteeing the quality and safety of medical services (4).

Given the distinct characteristics of the healthcare sector, hospital supply chains differ significantly from traditional supply chains in other industries. From a quality perspective, hospital supply chains emphasize maintaining high standards due to its direct bearing on patient safety and well-being. Any deviation in quality can lead to severe health risks. From a performance perspective, hospital supply chains prioritize timely delivery, inventory management, and the prevention of medication and medical equipment shortages. In contrast, traditional supply chains often prioritize cost-effectiveness and meeting market demands. These significant differences make hospital supply chains more in need of avoiding the generation of subjective biases compared to traditional supply chains. Such biases can impose significant financial strains on hospitals and carry profound, irreversible consequences for the quality and safety of medical services. For instance, selecting an inappropriate drug can compromise therapeutic outcomes or lead to severe side effects; opting for unsuitable medical equipment can increase surgical risks; and delays in the supply chain can cause shortages of crucial drugs or equipment, impacting urgent surgeries or treatments. These issues not only threaten patient health and safety but also risk damaging the hospital's reputation and may result in legal disputes. Regrettably, the internal control within public hospitals currently predominantly relies on the subjective decisions of managers, and the control schemes are usually determined by discussions among multiple individuals involved in its administrative management (5). This may result in deviations between certain decisions and actual needs, consequently impacting the operational efficiency of hospitals. Despite these issues, effective solutions are lacking. Therefore, the urgent task in public hospital supply chain management is to design and develop a method that can precisely and objectively evaluate and optimize the internal control of public hospital supply chains.

In recent years, a plethora of studies have been dedicated to exploring how to reduce subjective biases in hospital internal controls. Patil and Kant (6) propose a framework based on fuzzy analytical hierarchy process (AHP) and fuzzy technique for order performance by similarity to ideal solution (TOPSIS) to identify and rank the solutions of knowledge management adoption in supply chain and overcome its barriers; Samvedi et al. (7) proposed to use the analytical hierarchy process (AHP) and the fuzzy technique for order preference by similarity to the ideal solution (TOPSIS) to assess the risk in the supply chain. Venkatesh et al. (8) proposed to use the AHP-TOPSIS method to evaluate the supplier selection module in the humanitarian aid supply chain, thereby reducing the impact of subjective biases; Kumar and Singh (9) delved into the application of an integrated approach combining fuzzy Analytical Hierarchy Process (fuzzy AHP) and TOPSIS to evaluate the performance of global third-party logistics service providers for enhancing supply chain management effectiveness.

Previous studies primarily utilized either individual quantitative analysis or combined the Analytic Hierarchy Process (AHP) with the Technique for Order of Preference by Similarity to Ideal Solution (TOPSIS) to assess internal controls within supply chains, aiming to mitigate subjective biases. While TOPSIS can mitigate the issue of subjective bias to some extent and provide a structured decision-making framework, the reliance of this method on human judgment when determining criterion weights and evaluating alternative solutions may still introduce biases. These biases, stemming from personal perceptions, experiences, or cognitive inclinations, can jeopardize the assessment's reliability and validity, particularly when evaluators' judgments diverge. Recognizing these challenges, we aimed to bolster the evaluation's robustness. We incorporated the Game Theory Combination Weighting Method into our study's hospital internal control assessment of supply chains to counteract the inherent subjective biases of AHP. This method adeptly marries the strengths of AHP with the Entropy Weighting Method's objectivity, facilitating a more logical computation of indicator weights. It aligns decision-making subjectivity with each indicator's intrinsic value, ensuring the assessment remains uninfluenced by individual biases, thus enhancing its objectivity, precision, and depth, and producing more steadfast and credible results.

Based on the AHP-EWM (Analytic Hierarchy Process-Entropy Weighting Method) combined predictive model established by Chengguang et al. (10), we first embarked on a comprehensive acquisition and integration of internal control indexes within the public hospital supply chain. This was achieved through extensive literature analysis, questionnaire surveys, and expert interviews, thereby constructing a comprehensive internal control index system for public hospital supply chains. Then, we employed the Analytic Hierarchy Process and Entropy Weighting Method to derive both subjective and objective weights for each index. Subsequently, the Game Theory Combination Weighting Method was adopted to calculate the comprehensive weight of the indexes. Guided by the results of these weight computations, we innovatively proposed a series of internal control schemes for supply chains. These schemes were subsequently put into practice over a four-year period at the Weifang Maternal and Child Health Hospital. Following this implementation, we utilized the Fuzzy Comprehensive Evaluation Method to independently evaluate the hospital's internal control status within its supply chain, both before and after the introduction of our proposed design scheme. The Evaluation enabled us to test the reliability of the indicator system we established and to verify the feasibility of applying the Game Theory Combination Weighting Method to internal control within public hospital supply chains.

2. Method

2.1. Hierarchical analysis method

The Analytic Hierarchy Process (AHP) is a structured technique for complex decision-making, widely applied in areas like management, production, traffic safety, and user experience. It decomposes

decision-making objectives hierarchically, aiming to quantify and compare multiple projects and indices. The process involves:

1. Constructing a judgment matrix based on relative importance, as shown in Table 1 (11, 12).
2. Using binary comparison to assess related internal control indices at the same level. Managers score the index ratio based on a scale in Table 2, ranging from equal to extreme importance.
3. Engaging experts to compare and score indices at various levels, determining relative importance between criterion and sub-criterion layers, and deriving the judgment matrix. The eigenvector method is applied to the matrix, yielding the weight vector ω. The largest eigenvalue's eigenvector is normalized to determine each index's weight (13).

$$A\omega = \lambda_{\max}\omega$$

To account for evaluators' subjective biases, a consistency test is performed on the judgment matrix. This ensures reliable, consistent results and minimizes weight calculation uncertainty (14–16). The formulas used include:

$$M_{CI} = \frac{\lambda_{\max} - n}{n-1}$$

$$M_{CR} = \frac{M_{CI}}{M_{RI}}$$

Where λ_{max} is the maximum eigenvalue and n is the matrix order. If $M_{CR} > 0.1$, the matrix's weight distribution is deemed consistent; otherwise, it's contradictory.

2.2. Entropy weighting method

The entropy weighting method is a decision-making method that determines the weight of each index by comprehensively considering the amount of information provided by the measurement value of each index. This method takes into account the amount of information provided by each factor and calculates the information entropy of each index to measure its contribution to the overall system. The entropy weighting method is often employed as an approach for obtaining the objective weight, which involves determining the weight of each index based on the differences between the values of multiple indexes in the scheme. The specific steps are as follows:

Invite m experts to evaluate the n indexes, and the evaluation score ranges from 1 to 10. The higher the score, the more important the index is.

TABLE 1 Construction method of judgment matrix.

A	B_1	B_2	...	B_n
B_1	B_{11}	B_{12}	...	B_{1n}
B_2	B_{21}	B_{22}	...	B_{2n}
...	...	...		...
B_n	B_{n1}	B_{n2}	...	B_{nn}

Build an evaluation matrix B:

$$B = (b_{ij})_{m\times n} = \begin{bmatrix} b_{11} & \cdots & b_{1n} \\ \vdots & & \vdots \\ b_{m1} & \cdots & b_{mn} \end{bmatrix}$$

Normalize a matrix:

$$x'_{ij} = \frac{x_{ij}}{\sum_{i=1}^{m} x_{ij}}$$

Information entropy value of each index:

$$e_j = -\frac{1}{\ln m}\sum_{i=1}^{m} x'_{ij} \ln x'_{ij}$$

The information utility value of each index:

$$d_j = 1 - e_j$$

Get the objective weight of each index:

$$W_j = \frac{d_j}{\sum_{j=1}^{n} d_j}$$

2.3. Combination weighting method of game theory

Game theory, a branch of operations research, is concerned with the study of decision-making and strategic interactions among agents with conflicting interests. It is a mathematical framework that analyzes the incentives, strategies, and outcomes of situations where multiple participants make decisions. Game theory considers differences between expected and actual behaviors in weight calculations and seeks to identify optimal strategies for achieving desired outcomes. This approach ensures that the overall assessment remains unaffected by the subjective biases of individual evaluators, effectively addressing the inherent subjectivity pitfalls associated with the Analytic Hierarchy Process. The following is the method flow.

First, establish the basic weight vector set Wq = {ω1, ω2,..., ωn} ($q = 1, 2,..., p$). Among them, ω is the set of weights determined by the p weighting method, n is the number of indexes, and p is the number of methods for obtaining weights. The comprehensive weight is calculated by the subjective weight obtained by the AHP method and the objective weight obtained by the entropy weighting method, so $p = 2$. Set $\alpha = \{\alpha 1, \alpha 2\}$ is the linear combination coefficient, then the linear combination of the subjective weight and the objective weight vector is:

$$W = \alpha_1\omega_1^T + \alpha_2\omega_2^T$$

Based on the idea of the game aggregation model, two linear combination coefficients are optimized to minimize dispersion to

TABLE 2 Scaling of judgment matrix.

Scale value	Importance	Meaning
1	Equally important	Both indexes are equally important
3	Slightly important	The former metric is slightly more important than the latter
5	Obviously important	The former metric is significantly more important than the latter
7	Strongly important	The former metric is strongly more important than the latter
9	Extremely important	The former metric is definitely more important than the latter
2, 4, 6, 8	Median	The median of the above adjacent judgments
1, 1/2,...,1/9	Reciprocal	To the same extent, the importance of the latter compared with the former

obtain the most satisfactory weight in W. The established objective function is:

$$\min \left|\left| \sum_{p=1}^{n} \alpha_p \omega_p{}^T - \omega_p \right|\right|$$

Transform the above equation into a system of linear equations that optimize the first derivative condition:

$$\begin{bmatrix} \omega_1\omega_1{}^T & \omega_1\omega_2{}^T \\ \omega_2\omega_1{}^T & \omega_2\omega_2{}^T \end{bmatrix} \begin{bmatrix} \alpha_1 \\ \alpha_2 \end{bmatrix} = \begin{bmatrix} \omega_1\omega_1{}^T \\ \omega_2\omega_2{}^T \end{bmatrix}$$

Normalize processing α_1、α_2:

$$\begin{cases} \alpha_1{}^* = \dfrac{\alpha_1}{\alpha_1 + \alpha_2} \\ \alpha_2{}^* = \dfrac{\alpha_2}{\alpha_1 + \alpha_2} \end{cases}$$

Calculate comprehensive weight:

$$W = \alpha_1{}^* \omega_1{}^T + \alpha_2{}^* \omega_2{}^T$$

2.4. Fuzzy comprehensive evaluation

The fuzzy comprehensive evaluation method is a multi-objective assessment technique rooted in fuzzy mathematics' membership degree theory. Often termed as fuzzy multi-objective result evaluation, it's been extensively applied in domains like education, management, medicine, and risk assessment. This method, comprising three stages–setting the scoring criteria, forming the fuzzy rating matrix, and defining the weight vector–effectively manages fuzzy uncertainties, bolstering evaluation accuracy and impartiality. The evaluation process involves determining the comment set V for the plan, assigning its value, establishing the comprehensive weight vector for index levels (17), and constructing the evaluation matrix for relevant sub-criteria. Experts are then engaged to score the target, with the membership degree of comments determined based on their frequency. The matrix is as follows:

$$N_m = \begin{pmatrix} X_{11} & \cdots & X_{1y} \\ \vdots & & \vdots \\ X_{x1} & \cdots & X_{xy} \end{pmatrix}$$

In the formula, m is the level of the weight vector, X is the membership degree of different comments, x is the number of indexes included in the m level, and y is the number of subsets of the comment set.

According to the evaluation results of the sub-criteria layer, the weight vector of the criterion layer to the design scheme is calculated, and m is the level of the weight vector:

$$n_m = W_m N_m$$

Construct the criterion-level evaluation matrix:

$$n = \begin{pmatrix} n_1 \\ \vdots \\ n_m \end{pmatrix} = \begin{pmatrix} W_1N_1 & \cdots & W_mN_1 \\ \vdots & & \vdots \\ W_1N_m & \cdots & W_mN_m \end{pmatrix}$$

Calculate the comprehensive evaluation weight vector:

$$H = W_m n$$

Based on this, the total score of the relevant construction evaluation is obtained:

$$P = HV$$

3. Case study

In the process of evaluating internal control indexes, evaluators often have personal subjective judgments. To improve the objectivity and reduce uncertainty in the evaluation of internal control indexes, this study listed 12 specific contents related to the internal control business level of public hospitals based on the "Administrative Measures for Internal Control of Public Hospitals" issued by the National Health Commission of China in December 2020 (18). These contents were combined with the businesses included in the hospital supply chain, and internal control indexes were obtained through questionnaires, expert interviews, and literature analysis. Using the game theory combination weighting method, the weights of the internal control indexes of the public hospital supply chain were constructed. The research process is presented in Figure 1.

3.1. Index system construction

Given the intricate nature of the internal control of public hospital supply chains, it is essential to establish a comprehensive

and systematic set of indexes. To this end, this study reviewed pertinent literature on supply chains and internal control, consulted with internal control experts, and focused on the structure and processes of public hospital supply chains to identify the necessary internal control measures. To comprehensively analyze and summarize each link, the standard layer was divided into five levels, namely planning, purchasing, asset, expenditure business, and contract management. Through literature analysis, questionnaire survey, and expert interviews, the internal control indexes of the supply chain were obtained. Twenty six indexes were obtained through literature analysis (19, 20); 17 indexes were obtained through questionnaire surveys, and 19 indexes were obtained through expert interviews, for a total of 62 indexes. The indexes obtained through the three methods have a certain degree of repetition. To enhance the coherence and consistency of the index system, this study combined indexes with similar or identical meanings. For instance, "budget execution variance rate" and "budget analysis and assessment" were grouped under the inclusive relationship and referred to collectively as the index of "Budget Analysis and Assessment." The other indexes were merged in a similar way, and ultimately, a set of 17 indexes were obtained, constituting the internal control index system of the public hospital supply chain, as illustrated in Figure 2. To facilitate comparison, a brief description of each index is presented in Table 3.

The indicators are specifically designed for a wide array of public hospitals, ensuring their universal applicability, especially to tertiary hospitals at the prefecture and city level. However, due to the unique scope of indicators, special considerations are essential for some specialized hospitals. Nevertheless, the weighting method is universally applicable, suitable not only to various levels of public hospitals but also to specialized hospitals. This is attributable to the ability to employ the same method for weighting, despite the differing focal specialties of various hospitals.

3.2. Calculation results of subjective weights based on AHP

To establish the internal control index system for the public hospital supply chain, a panel of 10 experts in hospital supply chain internal control were invited to compare and score the 17 sub-criteria level indexes under the 5 categories. The purpose was to determine the standard layer and sub-criteria layer indexes. The relative importance between indexes was obtained, and the corresponding judgment matrix was constructed. The selection of the 10 experts was predicated on a set of predefined criteria, emphasizing their experience in hospital supply chain management, their academic pedigree, and their publication record within the domain of hospital supply chain management. Among these experts, four held doctoral degrees in supply chain management or related fields. Seven of them had dedicated over a decade to hospital supply chain management, and half were distinguished members of the China Association for Health Economics Management. The diversity and extensive expertise of this panel were instrumental in furnishing our research with in-depth and comprehensive insights, bolstering its robustness and credibility.

To effectively mitigate the subjective bias inherent in AHP, we adopted a multi-frequency expert scoring strategy. Specifically, we invited 10 experts to score multiple times and asked them to select the score they were most satisfied with from all their evaluations as the final score. Additionally, to further ensure the accuracy and consistency of the evaluations, we conducted a consistency test to ensure that all expert scores were within an acceptable range, thereby minimizing the impact of subjective bias to the greatest extent possible.

The evaluation opinions of 10 experts were integrated using the arithmetic mean and the AHP method was employed to analyze the internal control indexes of the public hospital supply chain. As a result, the subjective weight vector set ω_1 was obtained, which is presented in Table 4. All data have passed the consistency test, ensuring the reliability of the evaluation process.

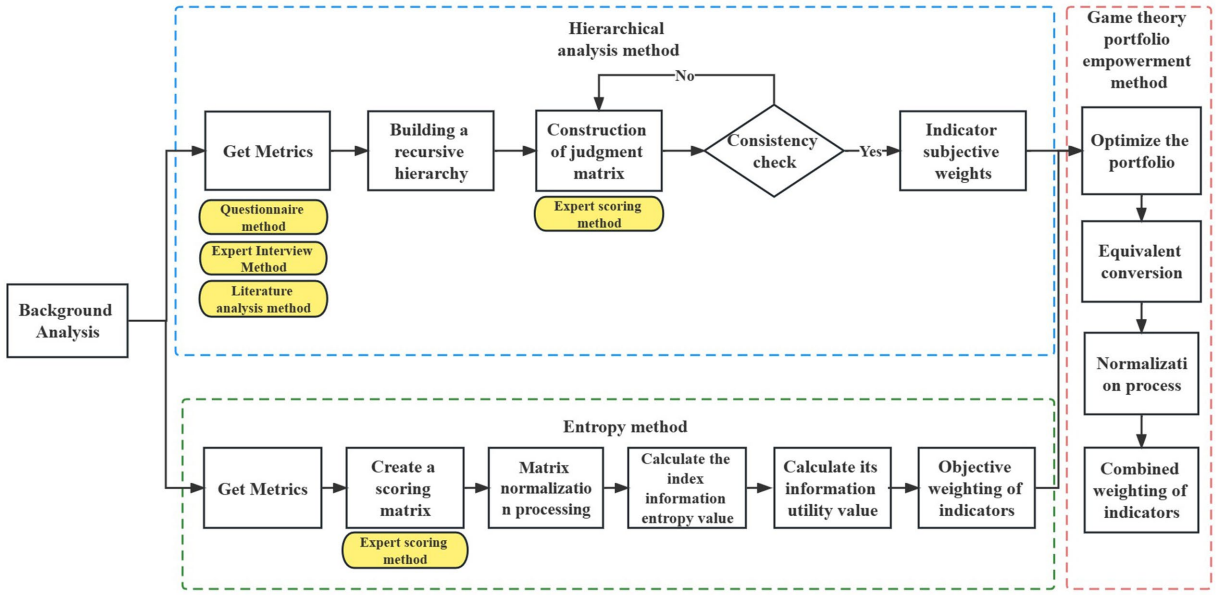

FIGURE 1
The process of constructing the internal control index system of the supply chain based on combination weighting method of game theory.

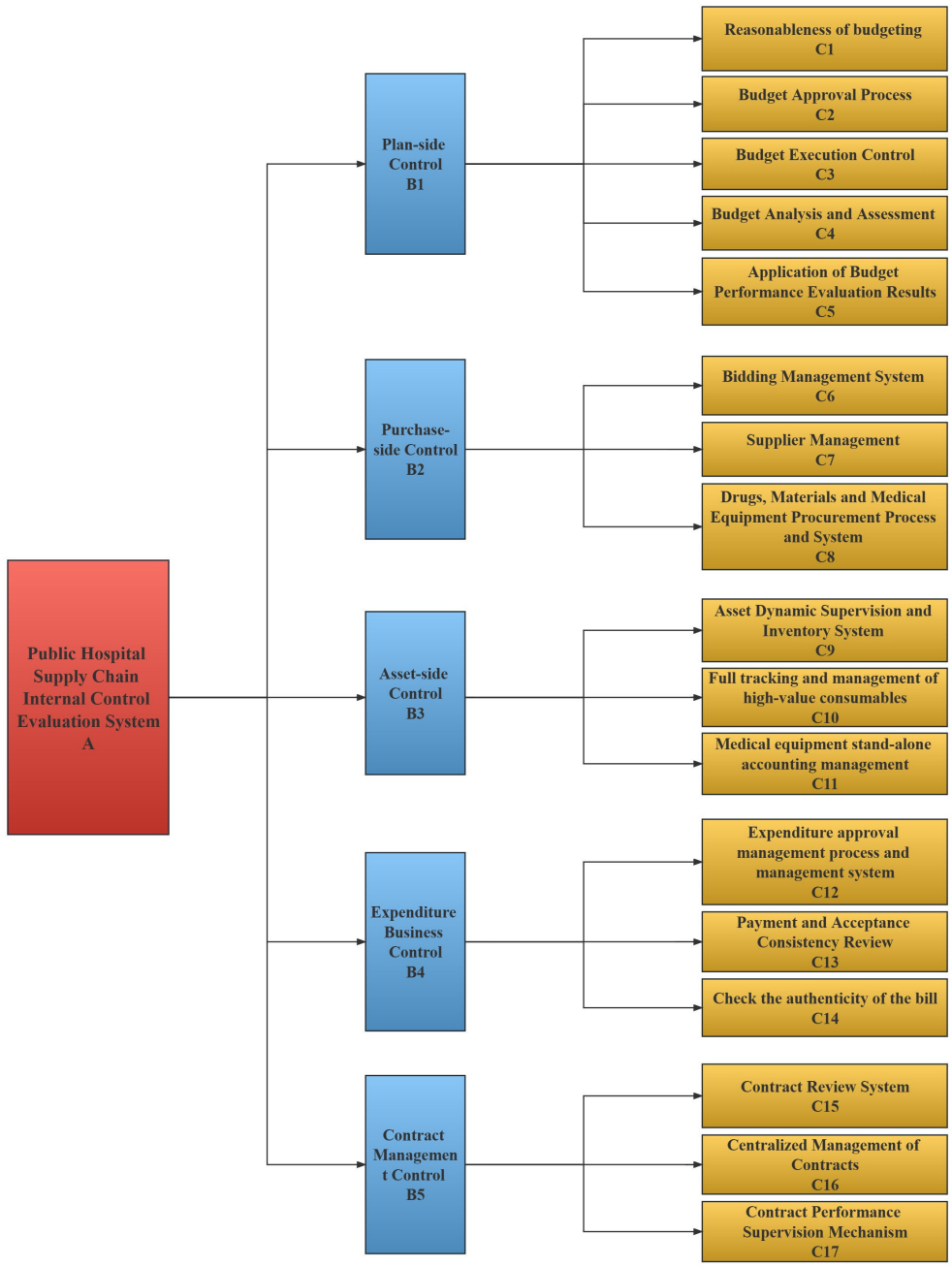

FIGURE 2
Internal control index system of public hospital supply chain.

3.3. Objective weight calculation results based on entropy weighting method

Six experts were invited to score the criteria and sub-criteria level indexes of the supply chain internal control. To ensure the objectivity and accuracy of the evaluation data, we adhered to the same selection criteria as before and chose an additional six experts who were not part of the original panel. Among these experts, two hold doctorate degrees in supply chain management or related fields. Five of them have dedicated over 10 years to hospital supply chain management, and all six are distinguished members of the China Health Economics Association.

The scoring results were used to construct an evaluation matrix (see Table 5 for details), from which the objective weight vector set ω2 of each index was obtained using the entropy weighting method calculation formula. The obtained objective weight vector set ω_2 is shown in Table 6.

3.4. Comprehensive weight calculation results based on game theory combination weighting method

By combining the subjective and objective weight calculation results and applying the relevant formula of the game theory

TABLE 3 Summary of indexes.

First-level index	Second-level Index	Index description
Plan-side control	Reasonableness of budgeting	The compilation method is scientific, and the content is comprehensive, which is in line with the actual development of the hospital
	Budget Approval Process	Rigorous approval procedure, perfect process, approval step by step, separation of incompatible positions, efficient and in line with internal control requirements
	Budget execution control	There are corresponding control methods, the execution is related to the budget, no budget is not purchased, no budget is not spent
	Budget analysis and Assessment	Regularly analyze the variance rate of budget execution, establish a budget analysis and assessment system, and design corresponding assessment indexes
	Application of Budget Performance Evaluation Results	Follow up the completion of performance goals promptly, with reasonable reward and punishment measures
Purchase-side control	Bidding management system	The bidding management system is sound, the process is perfect, and there are corresponding control measures
	Supplier management	Two-way control of procurement and supplier collaboration, supervision of supplier qualification certificates
	Drugs, Materials and Medical Equipment Procurement Process and System C8	Establish and improve the procurement system and process of drugs, materials, and medical equipment, have corresponding control measures and separate incompatible positions from each other
Asset-side control	Asset dynamic supervision and inventory system	Dynamically grasp the storage and use of assets, and establish a regular inventory system
	Full tracking and management of high-value consumables	The whole process of approval and tracking management is implemented from application for access to use to realizing the write-off of doctor's orders, and to support departments to provide feedback on the use of high-value consumables
	Medical equipment stand-alone accounting management	Realize one-machine-one accounting for key equipment, and evaluate equipment utilization efficiency
Expenditure business control	Expenditure approval management process and management system	Establish an expenditure approval process and system, which is related to the budget, no budget is not spent, and no budget is exceeded.
	Payment and acceptance consistency review	The payment business is associated with the storage document, one single code, to prevent repeated payment and wrong payment
	Check the authenticity of the bill	Validation and review of payment receipts
Contract management control	Contract review system	Establish a joint review system and process to jointly supervise the conclusion of contracts by multiple departments
	Centralized management of contracts	Set up an operation department to manage economic business contracts in a unified manner
	Contract performance supervision mechanism	Supervise the performance of the contract and ensure the consistency of contract execution and terms

combination weighting method, the comprehensive weight of each index was calculated using the game combination weighting method. The comprehensive weight of the criterion layer indexes was further calculated to construct the reference standard of the evaluation plan, which is presented in Table 7.

Leveraging the previously derived weight results, this study conceived a robust scheme for managing internal control within public hospital supply chains. This scheme was brought to life at the Weifang Maternal and Child Health Hospital, serving as a practical testament to the proposed concept. The effectiveness of this real-world application was then scrutinized through a fuzzy evaluation method, providing a comprehensive assessment of its performance. What follows is a detailed exposition of the modifications made to the scheme and the outcomes gathered from its practical deployment.

3.5. Scheme description and implementation details

Weifang Maternal and Child Health Hospital (referred to as F Hospital) is a specialized public hospital of tertiary first-class level with a development history of over 60 years. Guided by the weightings of various indicators provided by the combination weighting method based on game theory, we have implemented the following improvements to the internal control management of the hospital's supply chain:

To enhance control on the planning side, we introduced a centralized management system for all functional departments. We concurrently adopted an adaptive management system that supports a variety of procurement strategies, tailored to meet the needs of diverse stakeholders. This innovative system ensures that

procurement processes closely align with the real-time demands of our operations. To enhance control on the procurement side, we instituted a comprehensive supplier management protocol. This new system meticulously governs the process of supplier selection, evaluation, and performance management, ensuring consistency and high standards throughout the supply chain. We made substantial use of a Hospital Resource Planning (HRP) system to link bidding procedures with our established budgets, which fostered a unique synergy between these two previously independent processes. This integration allowed us to achieve an unprecedented level of proactive control over the bidding process. Furthermore, we introduced an early warning system for inventory control, notifying key personnel of both minimum and maximum stock thresholds. This proactive approach not only ensures smooth operations but also promotes economic efficiency by maintaining optimal inventory levels. Regarding the control on the asset side, we employed the logistics and fixed assets modules of the HRP system, leading to the establishment of secondary warehouses in clinical departments. This system transformation enabled the hospital to enact effective chargeable materials write-off policies, manage non-chargeable materials quotas, and maintain comprehensive traceability management for high-value consumables. As for contract management, we streamlined the process by placing hospital procurement contracts under the centralized control of the operations management department. The contract counter-signing process was seamlessly integrated into the HRP system. This approach empowered different departments, such as bidding, auditing, finance, discipline inspection, and legal affairs, to effectively perform their roles in the counter-signing process. Moreover, it ensured complete transparency and accessibility of every phase of contract execution within the system.

TABLE 4 Subjective weights of indexes.

Index	B1	B2	B3	B4	B5	Subjective weight
A	0.1762	0.3016	0.2236	0.1710	0.1276	
C_1	0.2314					0.0408
C_2	0.2076					0.0366
C_3	0.3452					0.0608
C_4	0.0927					0.0163
C_5	0.1231					0.0217
C_6		0.3024				0.0912
C_7		0.1938				0.0585
C_8		0.5038				0.1519
C_9			0.4328			0.0968
C_{10}			0.3796			0.0849
C_{11}			0.1876			0.0419
C_{12}				0.5917		0.1012
C_{13}				0.2784		0.0476
C_{14}				0.1299		0.0222
C_{15}					0.3961	0.0505
C_{16}					0.1716	0.0219
C_{17}					0.4323	0.0552

3.6. Comprehensive evaluation based on fuzzy evaluation method

As of now, the scheme outlined above has been in a state of continuous refinement and implementation for close to 4 years, spanning from September 2019 to April 2023. In this research, we have made use of fuzzy evaluation techniques to perform a holistic comparison. This includes a detailed appraisal of the internal supply chain control system within Hospital F as it was in 2019 and its subsequent transformation following the implementation of our proposed scheme in 2023. The specifics of this evaluation process are elaborated in the following sections.

3.6.1. Comprehensive evaluation of supply chain internal control in F hospital in 2019

First, determine the comment set $V=\{$very satisfied, satisfied, average, dissatisfied, very dissatisfied$\}$for the internal control scheme of the supply chain and assign it $V=\{100, 80, 60, 40, 20\}$.

Second, determine the comprehensive weight vector of indexes at different levels (18). According to the comprehensive weights of the indexes of the game combination weighting above, the comprehensive weight vector of the criterion layer is $W_A=(0.096668, 0.060498, 0.145919, 0.026494, 0.029992, 0.058053, 0.030089, 0.223408, 0.069657, 0.042869, 0.021269, 0.058079, 0.025208, 0.029993, 0.030068, 0.026509, 0.025228)$.

Third, to construct the fuzzy comprehensive evaluation matrix of the sub-criteria level of the internal control index of the F Hospital supply chain, we invited the original 10 experts who evaluated the indicators to assess and score the supply chain internal control construction of F Hospital. The experts determined the relevance of evaluation comments based on their frequency. The matrices N1 to N5 represent the comprehensive evaluations for plan-side control, purchase-side control, asset-side control, expenditure business control, and contract management control.

TABLE 5 Expert scoring results.

Expert	C_1	C_2	C_3	C_4	C_5	C_6	C_7	C_8	C_9	C_{10}	C_{11}	C_{12}	C_{13}	C_{14}	C_{15}	C_{16}	C_{17}
1	8	7	9	5	6	8	6	10	9	7	5	8	7	6	6	5	7
2	8	7	8	5	6	7	5	8	8	7	5	8	7	5	6	5	7
3	6	6	7	5	5	6	6	6	7	7	6	6	6	6	6	5	6
4	6	7	5	5	6	6	6	5	7	6	5	7	6	6	6	4	7
5	7	6	7	4	6	8	6	7	8	6	5	6	7	6	5	5	6
6	9	5	6	5	5	7	5	9	6	8	5	7	6	5	5	5	6

TABLE 6 Objective weights of design indexes.

Index	Information entropy	Degree of difference	Weights
C_1	0.99363699	0.00636301	0.096815241
C_2	0.996019759	0.003980241	0.060560637
C_3	0.990394973	0.009605027	0.146143568
C_4	0.998256948	0.001743052	0.026521103
C_5	0.998027399	0.001972601	0.030013757
C_6	0.996190309	0.003809691	0.057965676
C_7	0.998027399	0.001972601	0.030013757
C_8	0.985304501	0.014695499	0.223596725
C_9	0.995426608	0.004573392	0.069585628
C_{10}	0.99718981	0.00281019	0.042757948
C_{11}	0.998605717	0.001394283	0.021214469
C_{12}	0.996190309	0.003809691	0.057965676
C_{13}	0.998347153	0.001652847	0.025148599
C_{14}	0.998027399	0.001972601	0.030013757
C_{15}	0.998027399	0.001972601	0.030013757
C_{16}	0.998256948	0.001743052	0.026521103
C_{17}	0.998347153	0.001652847	0.025148599

$$N_1 = \begin{pmatrix} 0.6 & 0.3 & 0.1 & 0 & 0 \\ 0.5 & 0.4 & 0.1 & 0 & 0 \\ 0.3 & 0.4 & 0.3 & 0 & 0 \\ 0.2 & 0.4 & 0.4 & 0 & 0 \\ 0 & 0.4 & 0.4 & 0.2 & 0 \end{pmatrix}$$

$$N_2 = \begin{pmatrix} 0.3 & 0.5 & 0.2 & 0 & 0 \\ 0 & 0.5 & 0.4 & 0.1 & 0 \\ 0.6 & 0.4 & 0 & 0 & 0 \end{pmatrix}$$

$$N_3 = \begin{pmatrix} 0.4 & 0.5 & 0.1 & 0 & 0 \\ 0.8 & 0.2 & 0 & 0 & 0 \\ 0.2 & 0.6 & 0.2 & 0 & 0 \end{pmatrix}$$

$$N_4 = \begin{pmatrix} 0.9 & 0.1 & 0 & 0 & 0 \\ 0.6 & 0.3 & 0.1 & 0 & 0 \\ 0.1 & 0.6 & 0.3 & 0 & 0 \end{pmatrix}$$

$$N_5 = \begin{pmatrix} 0.5 & 0.5 & 0 & 0 & 0 \\ 0.3 & 0.6 & 0.1 & 0 & 0 \\ 0 & 0.5 & 0.4 & 0.1 & 0 \end{pmatrix}$$

Based on this, the total score of F Hospital supply chain internal control construction can be obtained:

$$P = \mathrm{H}V = 85.69$$

TABLE 7 Composite weighting results.

Index	Analytic hierarchy process results	Entropy weighting method results	Game theory Combination weighting method results
C_1	0.0408	0.09681524	0.096668
C_2	0.0366	0.06056064	0.060498
C_3	0.0608	0.14614357	0.145919
C_4	0.0163	0.0265211	0.026494
C_5	0.0217	0.03001376	0.029992
C_6	0.0912	0.05796568	0.058053
C_7	0.0585	0.03001376	0.030089
C_8	0.1519	0.22359673	0.223408
C_9	0.0968	0.06958563	0.069657
C_{10}	0.0849	0.04275795	0.042869
C_{11}	0.0419	0.02121447	0.021269
C_{12}	0.1012	0.05796568	0.058079
C_{13}	0.0476	0.0251486	0.025208
C_{14}	0.0222	0.03001376	0.029993
C_{15}	0.0505	0.03001376	0.030068
C_{16}	0.0219	0.0265211	0.026509
C_{17}	0.0552	0.0251486	0.025228

3.6.2. Comprehensive evaluation of supply chain internal control in F hospital in 2023

Building upon the assessment conducted in 2019, a subsequent evaluation of the supply chain internal control system was undertaken at F Hospital in April 2023. The evaluation aimed to measure the efficacy and continued improvement of the internal control scheme over a span of 4 years, as well as to identify areas that could be further optimized. A holistic comparison, employing the same fuzzy evaluation techniques, was utilized to accurately gage the evolution of the internal control system.

Similar to the 2019 evaluation, an expert panel of 10 supply chain internal control specialists was convened to conduct the evaluation and score the supply chain internal control construction of F Hospital. Subsequently, the fuzzy comprehensive evaluation matrix for each criterion level was generated (N_1-N_5).

$$N_1 = \begin{pmatrix} 0.8 & 0.2 & 0 & 0 & 0 \\ 0.7 & 0.3 & 0 & 0 & 0 \\ 0.6 & 0.3 & 0.1 & 0 & 0 \\ 0.6 & 0.3 & 0.1 & 0 & 0 \\ 0.1 & 0.6 & 0.2 & 0.1 & 0 \end{pmatrix}$$

$$N_2 = \begin{pmatrix} 0.5 & 0.4 & 0.1 & 0 & 0 \\ 0.2 & 0.6 & 0.2 & 0 & 0 \\ 0.7 & 0.3 & 0 & 0 & 0 \end{pmatrix}$$

$$N_3 = \begin{pmatrix} 0.6 & 0.3 & 0.1 & 0 & 0 \\ 0.8 & 0.2 & 0 & 0 & 0 \\ 0.3 & 0.6 & 0.1 & 0 & 0 \end{pmatrix}$$

$$N_4 = \begin{pmatrix} 0.9 & 0.1 & 0 & 0 & 0 \\ 0.7 & 0.2 & 0.1 & 0 & 0 \\ 0.2 & 0.6 & 0.2 & 0 & 0 \end{pmatrix}$$

$$N_5 = \begin{pmatrix} 0.7 & 0.3 & 0 & 0 & 0 \\ 0.5 & 0.4 & 0.1 & 0 & 0 \\ 0.1 & 0.6 & 0.2 & 0.1 & 0 \end{pmatrix}$$

Based on the resultant evaluation matrices, the total score of F Hospital's supply chain internal control construction for 2023 was computed and is given by:

$$\mathrm{P} = \mathrm{HV} = 90.75.$$

This score, higher than the previous score of 85.69 obtained in 2019, underscores the successful refinement and implementation of the internal control scheme. The assessment showed an increased satisfaction level across all criteria, signifying the effectiveness of the continuous improvement approach employed in F Hospital's supply chain internal control system. It is important to note, however, that this system is not static, and continued refinement will further enhance its effectiveness in the coming years.

4. Discussions

After evaluations in 2019 and 2023, a significant enhancement was observed in the internal control system of F Hospital's supply chain, evidenced by the score increase from 85.69 to 90.75. This change is not merely a superficial increase in numbers; it reflects profound real-world implications. Firstly, this growth signifies that during this period, we successfully optimized key processes and operations, thereby enhancing efficiency and output. Secondly, the rise in scores also indicates a notable increase in the satisfaction levels of both employees and patients, suggesting that our service quality has been acknowledged and appreciated. Most importantly, this consistent positive trend offers us a clear direction, validating that our investments in research and development are on the right track, laying a solid foundation for future strategic planning and decision-making.

During the pre-implementation and post-implementation phases of the scheme, we noted that core elements like the hospital's infrastructure, staff size, market trends, policy adjustments, technological advancements, and supplier relationships remained relatively stable over the years. This indicates that the increase in scores was not due to changes in other external conditional variables. Importantly, areas directly associated with the scheme witnessed a pronounced score increment, while those not emphasized in the scheme experienced only modest growth. Based on these observations, we have substantial grounds to believe that the notable score increase is intrinsically linked to the refinements made according to the proposed scheme. The score growth over these 4 years not only validates the accuracy of the metrics used in formulating the scheme but also accentuates the efficacy of the internal control design we instituted. The significant progress made within this period provides compelling evidence for the practicality and feasibility of the game theory combination weighting method in the realm of hospital supply chain internal control. This approach not only offers a balanced and precise assessment of the internal control system but also paves the way for the development of a more refined and scientifically rigorous control scheme.

In conclusion, the application of the game theory combination weighting method in designing hospital supply chain internal control systems has been empirically validated in F Hospital. This approach not only fortifies the scientific rigor of the control scheme design but also guarantees its sustained optimization and effectiveness. However, it's imperative to note that the findings of this study, being based solely on data from a single hospital, might have inherent limitations in terms of generalizability and applicability across diverse hospital settings. This suggests that the proposed method may require further validation and adjustments in different hospital contexts. Despite these potential constraints, the method can still be regarded as an innovative and reliable tool for the design of internal control mechanisms within public hospital supply chains.

5. Conclusion

This study presents a sophisticated approach to the enhancement of public hospital supply chain management by developing an internal control index system grounded in the game theory combination weighting method. This novel method addresses the issue of index duplication and augments the comprehensiveness and accuracy of the system. By transcending the constraints of traditional top-down designs, it encourages a more scientific, methodical approach to managing the supply chain internal control. This study utilizes Hospital F as a practical subject, employing the Fuzzy Evaluation Method to corroborate the feasibility of applying the Game Theory Combination Weighting Method in the context of hospital supply chain internal control. By evaluating the hospital before and after the implementation of the proposed design scheme, the effectiveness of the combined approach can be ascertained. The insights gleaned from this analysis can furnish valuable methodologies and pathways for future enhancements of internal control within hospital supply chains. Moreover, these findings can contribute to the broader advancement of supply chain management within the healthcare industry.

Despite the meaningful strides made in this research, we acknowledge its limitations and the need for further investigation. Firstly, the intricacy of supply chains and internal control systems warrants additional exploration to ascertain the comprehensiveness of the proposed internal control index system. Secondly, while we employed the Entropy Weighting Method to mitigate the influence of subjective biases in the Analytic Hierarchy Process, the weighting and evaluation results are still susceptible to expert preferences. This inherent bias is inevitable and must be considered. Thirdly, our findings are derived from a singular case study, which may not encompass the diversity and intricacy

of various hospital settings. Validation across multiple cases is essential to bolster the generalizability of our results. It should be emphasized that the proposed method and indicators for internal control within public hospital supply chains were designed with a broad perspective, aiming to cater to the general needs of public hospitals. However, individual hospitals, due to variations in their economic conditions and socio-cultural environments, may face unique challenges and requirements. For instance, specialty hospitals should consider their target audience and place emphasis on certain indicators accordingly. Therefore, while the foundational principles of the method are designed to be universally applicable, some customization and adjustments might be necessary based on the specific context and needs of each public hospital.

The future work will aim to address these limitations and further develop and refine the internal control index system. This study is an initial step toward a more comprehensive and scientifically robust evaluation of internal control systems within the hospital supply chain. The insights and outcomes from this research contribute to the ongoing discourse in this field and lay a foundation for future advancements.

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

This research was conducted following the ethical standards of the scientific community. All the data were collected with proper permissions, respecting confidentiality and privacy. Expert interviews were carried out with informed consent, and all practices respected the autonomy and rights of the participants. The study received appropriate ethical approval from relevant institutional bodies.

Author contributions

ZY conceptualized and designed the study, conducted the literature review, collected and analyzed the data, and wrote the manuscript and is accountable for all aspects of the work and ensures that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Conflict of interest

The 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.

Publisher's note

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Reporting reimbursement price decisions for onco-hematology drugs in Spain

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Introduction: Even using well-established technology assessment processes, the basis of the decisions on drug price and reimbursement are sometimes perceived as poorly informed and sometimes may be seen as disconnected from value. The literature remains inconclusive about how Health Technology Assessment Bodies (HTAb) should report the determinants of their decisions. This study evaluates the relationship between oncology and hematology drug list prices and structured value parameters at the time of reimbursement decision in Spain.

Methods: The study includes all new onco-hematological products (22), with a first indication authorized between January 2017 and December 2019 in Spain and pricing decisions published up until October 2022. For each product, 56 contextual and non-contextual indicators reflecting the structured multiple criteria decision analysis (MCDA) – Evidence-based Decision-Making (EVIDEM) framework were measured. The relationship between prices and the MCDA-EVIDEM framework was explored using univariate statistical analyses.

Results: Higher prices were observed when the standard of care included for combinations, if there were references to long-lasting responses, for fixed-duration treatment compared to treatment until progression and treatment with lower frequencies of administration; lower prices were observed for oral administration compared to other routes of administration. Statistically significant associations were observed between prices and the median duration of treatment, the impact on patient autonomy, the ease of use of the drug, and the recommendations of experts.

Discussion: The study suggests that indicators related to the type of standard of care, references to long-lasting responders, the convenience of the use of the drug, and the impact of treatment on patient autonomy, as well as contextual indicators such as the existence of previous clinical consensus, are factors in setting oncology drug prices in Spain. The implementation of MCDA-EVIDEM methodologies may be useful to capture the influence on pricing decisions of additional factors not included in legislation or consolidated assessment frameworks such as the European Network for Health Technology Assessment (EunetHTA) core model. It may be opportune to consider this in the upcoming revision of the Spanish regulation for health technology assessments and pricing and reimbursement procedures.

KEYWORDS

health technology assessment, multicriteria assessment methods, price and reimbursement systems, onco-hematologic prices, value assessment

1. Introduction

Concerns about the increasing cost of oncological and hematologic innovation in Europe are growing as prices of cancer drugs are high but not always related to a proportional improvement in patient health status (1). In Europe, the increase in the rate of health spending on cancer has been faster than the increase in cancer incidence during the last 20 years. Similarly, the loss of productivity related to premature cancer mortality has decreased, while productivity loss related to morbidity is still uncertain (2).

Progressively flexible regulatory criteria for authorization in the setting of precision medicine aims to accelerate market access decisions at the pricing and reimbursement process. Studies of authorization decisions in Europe have estimated that after monitoring post-authorization real-world evidence for 3.3 years, benefits on survival of those authorized drugs were only observed in 7% of cases, and improvement in reported quality of life was achieved in only 11% of them (3). A recent study (4) confirms that this trend is consolidated, and regulatory practice is biased toward earlier access at the expense of the production of post-authorization robust evidence, especially when the drug covers clinical unmet needs in diseases with poor prognosis (5). Pricing and reimbursement decisions are tough when evidence is scarce and lacking comparative data, risking opportunity costs (6). In order to minimize this, new access management models have been implemented across Europe during the last decades (7), although to a limited extent and with a lack of methodological harmonization (8). The increase in prices of oncologic products has generated additional international concerns (9) about the disconnection between price and value.

There is still an open debate in Europe about which are the adequate methods to assess the value of drugs (10). Methods of setting "fair prices" are generally focused on clinical benefits or expanded to the so-called value-based pricing, which is usually focused on cost-effectiveness analysis (11–13). Cancer drugs are normally classified as innovation based on implicit clinical value through Quality-Adjusted Life-Years – QALYS (e.g., United Kingdom, Australia, Sweden) or using innovation scales (e.g., Canada, Japan, France, Germany, Austria, Italy) (14). However, healthcare authorities do not normally unveil the details of the methodology applied to assess value, while new cancer drugs are increasingly reimbursed at a higher price than the available alternatives (15).

Recent studies (16) show that even in countries with well-established technology assessment processes (such as the UK, Germany, France, and Switzerland), prices may still be considered disconnected from value. In fact, in countries such as France, Australia, or the UK, prices are only weakly associated with drug clinical benefits (17–19).

Besides a lack of elements to check consistency between price and value, the literature remains inconclusive about the factors that Health Technology Assessment Bodies (HTAb) are using to make their decisions on value and how the payers are deciding and reporting price decisions, especially when applying managed entry agreements (20). Recent studies (21) show that EVIDEM's framework provides a complete and suitable value assessment framework, including contextual dimensions, and it has been progressively adopted by some HTAb in Europe. Additionally, differences may exist in the concept of value between payers and patients: while payers are generally focused on objective clinical outcomes to determine reimbursement conditions, the importance of patient preferences is not clear (22, 23).

In Spain, the pricing and decision process starts after the European marketing authorization is formally adopted by the Spanish Agency of Medicines and Medical Devices (AEMPS) (24). Subsequently, a Therapeutic Positioning Report (TPR) is issued by the REvalMed network (25) to inform about the added therapeutic value of the drug compared to current therapeutic alternatives. The TPR includes a therapeutic evaluation from the AEMPS, an economic assessment from the General Directorate for Common Portfolio of the NHS and Pharmacy Services (DGCCSF), and a final technical revision by external experts and scientific societies appointed by the REvalMed network. The TPR, together with the application dossier filed by the marketing authorization holder and DGCCSF's reports, is supposed to be the main driver for reimbursement decisions. The Inter-ministerial Committee on Pricing of Medicines and Healthcare Products (CIPM) is the body responsible for the final resolution of price and reimbursement conditions (26). The CIPM decision is published as a listed price (not net price) and motivation in general terms, which are based on the criteria listed in the RDL 1/2015, but the information provided by the Ministry of Health (MoH) is not detailed enough to know how the value of the drug has been established. It has been questioned whether the Spanish pricing model is based only on budgetary impact and lower European nominal price, without accounting for contextual criteria and societal needs.

Detailed information on how Spanish healthcare authorities define price and reimbursement conditions of new drugs is not available, and a lack of predictability, potentially leading to inconsistency between value and price, has been alleged (27, 28). The Royal Legislative Decree 1/2015 (RDL 1/2015) of the Law on

Abbreviations: AEMPS, Spanish Agency of Medicines and Medical Devices; CIPM, Inter-ministerial Committee on Pricing of Medicines and Healthcare Products; DGCCSF, General Directorate for Common Portfolio of the NHS and Pharmacy Services; EPAR, European Public Assessment Report; ESMO-MCBS, European Society of Medical Oncology-Magnitude of Clinical Benefit Scale; EUnetHTA, European Network for Health Technology Assessment; EVIDEM, Evidence and Value Impact on Decision-Making; HTAb, Health Technology Assessment Bodies; ICER, Incremental Cost-Effectiveness Ratio; MCDA, Multi-Criteria Decision Analysis; MoH, Ministry of Health; NHS, National Health Service; NICE, National Institute for Health and Care Excellence; OS, Overall Survival; PFS, Progression-Free Survival; QALYS, Quality-Adjusted Life-Years; TPR, Therapeutic Positioning Report.

Guarantees and Rational Use of Medicines and Health Products (29) lists only a restricted set of criteria to be used by the Spanish National Health System to establish prices of publicly funded medicines.

Based on recent data released by MoH (30), 90% of assessed oncologic medicines in Spain are publicly funded, with a listed price 15 times higher than the average price of new non-cancer-related drugs. By 2021, cancer drug costs represented 16.9% of the global pharmaceutical Spanish public budget, and the cost of cancer drugs at the hospital level has grown by 105.9% since 2016. The main objective of this study is to externally evaluate whether there is a relationship between the prices of oncology and hematology drugs and the evidentiary and contextual information available at the time of reimbursement decision in Spain by applying a structured assessment of parameters measuring drug value and to identify the most relevant criteria related to price decisions made by health authorities.

2. Materials and methods

All new chemical entities with a first EMA authorization for a single onco-hematologic indication between January 2017 and December 2019 were identified, and price and reimbursement decisions of the Spanish MoH, including the notified price and public funding authorization, were tracked based on the publicly available database Bifimed (31) and the resolutions published by the MoH up until the end of October 2022 (Supplementary Table 2).

For standardization and comparison purposes, a daily treatment cost based on notified prices was assigned following the Summary of Product Characteristics recommended posology for the studied indication. When the treatment duration was fixed, the cost was annualized. Products with a negative decision were assigned a price of zero; no other data imputation was applied.

For each product, a set of indicators from the MCDA-EVIDEM framework was used. A literature review was carried out to identify the indicators (32–36) for each MCDA-EVIDEM dimension (Table 1). The inclusion criteria for the review were articles published from January 2017 to December 2021 that included MCDA-EVIDEM-related indicators to assess onco-hematologic drugs as well as country legislation and HTAb official documents available in English or Spanish. The review did not include outdated documents. The indicators for each product were extracted from available European Public Assessment Report (EPAR), TPR (37), European Society of Medical Oncology-Magnitude of Clinical Benefit Scale (ESMO-MCBS) evaluations (38), National Institute for Health and Care Excellence (NICE) economic assessments (39), and freely available information from national and regional healthcare authorities (40). The indicators were informed by a stepwise approach including two independent reviewers for each product, and discrepancies were resolved through discussion. Public notified reimbursed prices per product (expressed as annual cost per treatment) were also included.

Continuous variables for MCDA-EVIDEM dimension indicators were expressed as mean ± standard deviation, and categorical variables were expressed as percentages.

To evaluate the relation between oncology and hematology treatment prices and MCDA-EVIDEM indicators at the time of reimbursement decision, univariate analyses were performed. For correlation analyses, categories were normalized, summaries were calculated by dimension, and prices were categorized by terciles where required. To compare variables, the non-parametric Mann–Whitney test was used for continuous variables and the Fisher exact test was used for categorical variables. Spearman's coefficients and 95% confidence intervals were calculated to assess correlations. The statistical definition was set at 5% two-tailed. The analysis was deemed exploratory, and, thus, no measures to account for multiplicity were applied.

3. Results

From January 2017 to December 2019, 24 oncological new chemical entities were granted a first indication marketing authorization in Europe. One product was excluded due to a conflict of interest in the team, and an adjuvant product for photodynamic therapy was deemed not suitable for the exercise (41) (Figure 1). Eventually, 22 products that aimed to treat 11 different tumors were analyzed. By October 2022, pricing and reimbursement had been granted for 18 products and denied for four products (Table 2). The most frequent indications were breast and lung cancer, and nine drugs had orphan designation (Table 2). Only two products had no therapeutic alternatives (in lung and agnostic indications) and roughly half of the products had targeted therapies as alternative options. Likewise, half of the treatments had an impact on patient autonomy (long intravenous administration, daycare admission), mostly in acute leukemia, lymphomas, melanoma, and neuroblastoma. Products for the treatment of melanoma, breast, neuroblastoma, and agnostic indications showed longer Progression Free Survival (PFS), observed and compared to control, over the median (14 months), and better Overall Survival (OS) vs. control was seen for products to treat leukemia and neuroblastoma. Most of the products were aimed at non-curative settings (19/22), with a moderate MCBS score (13/22 products under the score of 4) and low quality of evidence (17/22 products under a JADAD score of 3). Most did not require new healthcare service delivery routes (14/22) and were administered orally (15/22). Many had an Incremental Cost-Effectiveness Ratio (ICER) over the NICE threshold and were included in the NICE Cancer Drugs Fund (16/22), and most were related to cancers included in National or Regional Health Plans (18/22). More than half of the products (12/22) were explicitly recommended by expert consensus or included in clinical practice guidelines, while 4/22 products were explicitly not recommended (Table 1).

The univariate analysis (Tables 3, 4) showed significantly higher listed prices when the standard of care was combined treatments, if long-lasting responders were reported, and for several characteristics of the treatment: higher prices for fixed-duration compared to treatment until progression and treatment with lower frequencies of administration, and lower prices for oral administration compared to other routes of administration. There were significant correlations between price and the ease of use of the drug, the impact of treatment on patient autonomy, and the existence of recommendations by experts. Regarding summaries by dimensions, the only association with price values was observed for the "expert consensus/clinical practice guidelines recommendations" dimension that contained a single item.

TABLE 1 Description of MCDA-EVIDEM dimensions and metrics.

Dimensions and indicators	Metrics	Mean (SD) or %	(N)
Noncontextual			
Disease severity			
Speed tumor growth	*Time of duplication (months)*	*13.64 (19.61)*	*20*
% Metastasized	*Percentage of patients with metastasis at diagnosis*	*50% (40%)*	*22*
Expected survival 5-years	*Percentage of patients with expected survival ≥ 5 years*	*29% (25%)*	*22*
Physical function and general health	*Normalized Score of SF36 – EQ5D – EORTC QLC or C30*	*62.41 (21.72)*	*12*
Size of affected population			
Prevalence	*Cases per 10,000 inhabitants*	*23.83 (219.32)*	*22*
Incidence	*New cases per 10,000 inhabitants and year*	*27.06 (29.57)*	*22*
Unmet needs			
Treatment options	*Percentage with/without alternative treatment options*	*With: 90%* *Without: 9%*	*22*
Type of standard of care	*Percentage of chemotherapy/immunotherapy/directed agents/surgery/radio/combined/ others/none*	*Chemotherapy:21%* *Directed agents: 47%* *Combined:17%* *Others: 4%* *None: 9%*	*22*
Comparative effectiveness			
Progression-free survival	*Months (median) during which patients have not experienced disease progression*	*13.69 (7.83)*	*22*
Progression-free survival vs. *control*	*Difference in months (median) during which patients have not experienced disease progression* vs. *control*	*6.73 (4.59)*	*22*
Objective response rate (RECIST/MRD)	*Percentage of patients that experience complete response and partial response*	*0.55 (0.17)*	*19*
Objective response rate (RECIST/MRD) vs. *Control*	*Difference in percentage of patients that experience complete response and partial response* vs. *control*	*20% (14%)*	*14*
Complete response (RECIST/MRD)	*Percentage of patients that experience complete response*	*23% (27%)*	*20*
Complete response (RECIST/MRD) vs. *control*	*Difference in percentage of patients that experience complete response* vs. *control*	*9% (13%)*	*15*
Partial response (RECIST /MRD)	*Percentage of patients that experience partial response*	*33% (18%)*	*18*
Partial response (RECIST /MRD) vs. *control*	*Difference in percentage of patients that experience partial response* vs. *control*	*10% (7%)*	*13*
Long responders	*Percentage of patients mentioned as long responders*	*Yes: 9%* *No: 91%*	*22*
Overall survival	*Months (median) of treatment randomized to death*	*25.61 (16.43)*	*15*
Overall survival vs. *control*	*Difference in months (median) of treatment randomized to death* vs. *control*	*9.23 (13.25)*	*12*
Comparative safety and tolerability			
Any adverse event	*Percentage of patients experiencing an adverse event*	*97% (6%)*	*22*
Any adverse event vs. *control*	*difference in percentage of patients experiencing an adverse event* vs. *control*	*5% (10%)*	*16*
Non-fatal serious adverse events (>3)	*Percentage of patients experiencing an adverse event of grade 3 to 5*	*57% (26%)*	*16*
Non-fatal serious adverse events (>3) vs. *control*	*Difference in percentage of patients experiencing an adverse event of grade 3 to 5* vs. *control*	*15% (19%)*	*16*
Fatal adverse events (Grade 5)	*Percentage of patients experiencing an adverse event of grade 5*	*7% (7%)*	*21*
Fatal adverse events (Grade 5)	*Difference in percentage of patients experiencing an adverse event of grade 5* vs. *control*	*1% (5%)*	*16*
Dosage adjustment due to adverse events	*Mention (yes/no) of dosage adjustment due to adverse effects*	*Yes: 73%* *No: 14%* *Not relevant: 13%*	*22*
Treatment discontinuation due to adverse events	*Percentage of patients discontinuing treatment due to adverse events*	*14% (10%)*	*22*
Treatment discontinuation due to adverse events vs. *control*	*Difference in percentage of patients discontinuing treatment due to adverse events* vs. *control*	*8% (7%)*	*22*

(Continued)

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TABLE 1 (Continued)

Dimensions and indicators	Metrics	Mean (SD) or %	(N)
Median duration of treatment	*Months (median) of duration of treatment*	*21.27 (24.54)*	*17*
Other Indications (patients exposed)	*Number of potential patients for all indications (exposed population as reported in EPAR)*	*920.95 (665.65)*	*22*
Comparative patient-perceived health and patient-reported outcomes			
Quality of Life	*Normalized score of quality-of-life scale*	*0.06 (0.22)*	*14*
Impact on autonomy	*Mentioned (yes/no) disruption of daily activities due to delivery of treatment*	*Yes: 41%* *No: 59%*	*22*
Frequency of treatment (administration)	*Dose administration by unit of time*	*Once a month: 4%* *Twice a month: 4%* *Once a week: 4%* *Twice a week: 0%* *>Twice a week: 9%* *Once a day: 48%* *Twice a day: 17%*	*22*
Variable treatment guideline	*Mentioned (yes/no) treatment guidelines changes*	*Yes: 13%* *No: 68%*	*22*
Time of treatment	*Mentioned (fixed/up to progression/variable) time of treatment*	*Fixed: 17%* *Up to progress: 50%* *Other: 36%*	*22*
Easy to use, mode and set of administration	*Mentioned (oral/injection/intrathecal) way of administration*	*Oral: 68%* *Injection: 27%* *Intrathecal: 4%*	*22*
Combined chemotherapy	*Mentioned (with/without) combination with chemotherapy*	*With: 18%* *Without: 81%*	*22*
Magnitude of therapeutic benefit (*)			
Magnitude of clinical benefits MCBS	*Scale of MCBS*	*3.14 (0.77)*	*22*
Type of benefit			
Curative/Non-Curative	*Mentioned (curative/non-curative) clinical benefit*	*Curative: 18%* *Non-Curative: 82%*	*22*
Comparative cost consequences – cost of intervention			
NICE ICER > threshold	*Mentioned (yes/no) NICE ICER > threshold before any patient access scheme was in place.*	*NA: 4%* *Yes: 73%* *No: 23%*	*21*
NICE cancer fund	*Mentioned (yes/no) inclusion as a NICE Cancer Fund's Drug*	*Yes: 36%* *No: 64%*	*22*
ICER (NICE value)	*Δ monthly target therapy cost / Δ time to disease progression as per NICE information*	*52,363.9 (28,859.4)*	*18*
Comparative cost consequences – other medical costs			
Cost treatment (procedures and tests-physician visits-hospitalizations…)	*Yearly direct medical costs (€) excluding purchasing costs of the technology (i) concomitant medications, (ii) outpatient visits, diagnostic/laboratory tests, hospitalizations, and other monitoring costs (including management AEs), and (iii) terminal care.*	*NA: 50%* *0: 45%* *>0: 5%*	*11*
Comparative cost consequences – non-medical costs			
Cost treatment	*Yearly cost of (€) treatment (based on notified prices)*	*NA: 100%*	*0*
Quality of evidence (**)			
JADAD scale	*JADAD scale*	*2.50 (1.40)*	*22*
Expert consensus and clinical practice guidelines			
Recommendation by experts	*Mentioned (yes/no) recommendation included in consensus available at the time of pricing*	*Recommended: 24%* *Not recommended: 76%*	*17*

(Continued)

TABLE 1 (Continued)

Dimensions and indicators	Metrics	Mean (SD) or %	(N)
Contextual			
Mandate and scope of the healthcare system			
Included in National/Sub-National Health Plan	*Type of cancer mentioned (yes/no) in healthcare plans*	*Included: 82%* *Not included: 18%*	*22*
Population priorities and access			
Preferences of the population as a need	*Type of cancer mentioned (yes/no) in official positions or documents from NGO's and Patient Advocacy Groups*	*Identified: 18%* *Not identified: 82%*	*22*
Common goal and specific interests			
Stakeholders' expression of interest and alignment	*Type of cancer mentioned (yes/no) in societal sources (mass or digital media)*	*Identified: 23%* *Not identified: 77%*	*22*
Environmental impact			
Impact of the intervention on environment – packaging, production	*Relevant environmental impact mentioned (yes/no) in EPAR*	*Yes: 21%* *No: 79%*	*19*
System capacity and appropriate use of intervention			
Healthcare services delivery change	*Mentioned (yes/no) change in healthcare service delivery or inversion (e.g., new biomarkers) to deliver care*	*Yes: 36%* *No: 64%*	*22*
Political, historical, or cultural context			
Societal acceptability of the decisions	*Type of cancer mentioned (yes/no) at legal level or included in political statements*	*Identified: 9%* *Not identified: 91%*	*22*

(*) Non-curative indications range from 1 (lowest) to 5 (highest) benefit. Curative indications range from A to C (A equalized to 5 and C to 1). (**) JADAD scores range from 0 (lowest) to 5 (highest) quality of trials.

FIGURE 1
Product selection.

4. Discussion

Our findings suggest that the initial price of oncology and hematology products tends to be influenced (higher prices) only by a few variables: the type of standard of care, the reporting of long-lasting responders, the convenience of use of drugs, the impact on patient autonomy, a limited duration of treatment, and contextual indicators such as the existence of previous clinical consensus. None of the individual items for comparative efficacy, safety, or quality of life reached significance for price correlation. Attempts to summarize values by dimensions compared to descriptions of individual items did not improve the explanation of price differences. However, the lack of standardized metrics and harmonized interpretation of contextual indicators limits the interpretation of the results.

The main limitation to moving forward with more transparent and standardized drug pricing processes is the lack of shared convention about the definition of "price" as an expression of "value" (1). For example, concepts such as quality-adjusted life years (to standardize health gains) do not capture the social perception of health benefits when the life expectancy of diseases differs (42). Additionally, price-setting processes are conditioned by available and previous therapeutic alternatives, influencing prices of pharmaceutical innovation based on historical inertia and baseline costs of the disease for the system (43). Additionally, dose, posology, and treatment duration add complexity to the direct comparison of value-based prices of new drugs.

There is a diversity of standardized clinical outcomes (overall survival, progression-free survival, quality of life, and safety) that medical societies and European healthcare authorities (38) are using to guide or define reimbursement conditions of oncology drugs (44).

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TABLE 2 Price and funding decisions by October 2022 for oncological products with first regulatory authorization* from January 2017 to December 2019.

Active principle	Indication	Date authorization	Date final P&R decision	Public funding	Time# to final P&R decision (days)	Yearly treatment cost ~ (public listing price)
Inotuzumab ozogamicin	Acute lymphoblastic leukemia	21/07/2017	1/7/2019	yes	710	189,431.35 €
Dinutuximab beta	Neuroblastoma	06/09/2018	1/6/2022	yes	1,364	171,998.95 €
Mogamulizumab	Squamous cell carcinoma	05/06/2019	1/7/2021	yes	757	160,158.35 €
Polatuzumab vedotin	Acute myeloid leukemia	18/02/2020	1/9/2021	yes	561	139,200.05 €
Brigatinib	Lung cancer	28/11/2019	1/5/2021	yes	520	109,781.05 €
Durvalumab	Lung cancer	31/10/2018	1/1/2020	yes	427	98,550.00 €
Rucaparib	Breast cancer	10/05/2019	1/1/2020	yes	236	91,129.55 €
Midostaurin	Chronic myelogenous leukemia	30/10/2017	1/4/2019	yes	518	86,997.75 €
Encorafenib	Melanoma	04/10/2018	1/9/2019	yes	332	86,844.45 €
Binimetinib	Melanoma	19/10/2018	1/9/2019	yes	317	86,844.45 €
Niraparib	Ovarian cancer	08/03/2018	1/8/2019	yes	511	64,918.90 €
Lorlatinib	Lung cancer	20/06/2019	1/2/2021	yes	592	63,630.45 €
Neratinib	Breast cancer	07/01/2020	1/7/2022	no	906	61,320.00 €
Ribociclib	Breast cancer	04/09/2017	1/11/2017	yes	58	57,936.45 €
Tivozanib	Renal cancer	09/04/2018	1/3/2019	yes	326	47,650.75 €
Abemaciclib	Breast cancer	26/10/2018	1/5/2019	yes	187	46,668.90 €
Citarabine/ daunorubicin	Acute myeloid leukemia	19/12/2018	1/3/2022	yes	1.168	42,639.30 €
Gemtuzumab ozogamicin	Acute myeloid leukemia	25/05/2018	1/7/2019	yes	402	35,999.95 €
Dacomitinib	Lung cancer	23/05/2019	1/8/2020	yes	436	32,850.00 €
Talazoparib	Breast cancer	24/07/2019	1/8/2021	no	739	0.00 €
Gilteritinib	Acute myeloid leukemia	05/12/2019	1/6/2021	no	544	0.00 €
Larotrectinib	Agnostic indication	21/11/2019	1/4/2022	no	862	0.00 €

*Cemiplimab was excluded because of a conflict of interest; padeliporfin was excluded because the indication was an adjuvant for photodynamic therapy. ~ Cost calculated according to posology in the product information for the studied indication and annualized where required if fixed maximum length of treatment. Costs of 0.00 € reflect negative price and reimbursement decisions by October 2022. #Time from the date of European Marketing Authorization until inclusion in the national reimbursement listing; since negative decisions and successive resubmissions may occur until reimbursement is granted, it does not reflect the length of pricing and reimbursement procedure.

Other reports (17, 19–21, 23, 45) suggest that perceived additional therapeutic benefits based on weak variables (such as response rates) or perception of severity (when this is measured) may be driving oncology drug prices. In our data, these clinical variables and "hard" variables such as overall survival were not good pricing predictors. However, we observed higher prices for products reporting references to long-lasting responders. Furthermore, our research also shows that other intermediate indicators such as PFS, generally accepted as indicators of the capacity of a drug to cure or alter the natural history of the disease (46), were not strong predictors of prices either. The lack of consistent evidence based on long-term efficacy data or relative efficacy data of new drugs vs. frequently used drugs at the time of price negotiations does not seem to have any penalty on the price and reimbursement decisions in Spain. The study also suggests the influence of contextual indicators, such as the existence of expert consensus and the impact of the route of administration to patients, on setting prices.

Several limitations of the study should be considered. Firstly, only a few new oncology drugs authorized for a first indication were analyzed. The influence that multiple indications may have in price negotiations requires further analysis. Secondly, the value assessment was made by evaluators working in the context of payers of healthcare services, so that may not fully reflect the perspectives of pricing and reimbursement decision-making. Third, we did not calculate summary indicators or overall scores for MCDA-EVIDEM, as suggested by others (43), since the exercise aimed to verify whether a more transparent reporting of the criteria used for decisions may help all stakeholders to predict the key determinants of value and to support the expectations of manufacturers, the information given to the lay public, and the consistency of the decision-making by authorities. Finally, we did not run a systematic search of the literature using a diverse range of databases to identify all potential studies analyzing the relationship between prices and the MCDA-EVIDEM framework, and there is a scarcity of references available on methods and definitions for data extraction and analysis; therefore, we cannot exclude that our work may be influenced by publication biases.

Our work may provide a basis for some proposals in the context of upcoming regulations and changes in Health Technology Assessments. The new European regulation (47) states that inclusive

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TABLE 3 Description of the mean (SD) listed yearly prices of oncology drugs according to the values of MCDA categorical items.

Variables and values	Mean	SD	Lower limit 95% CI	Upper limit 95% CI
Alternative treatment options				
With	78,800.06 €	52,983.23 €	55,308.61 €	131,783.30 €
Without	49,275.00 €	69,685.37 €	18,378.24 €	80,171.76 €
Type of standard of care				
Chemotherapy	68,353.67 €	66,410.51 €	38,908.90 €	97,798.44 €
Combined	143,946.84 €	43,194.75 €	124,795.36 €	163,098.32 €
Directed agents	59,858.69 €	29,376.64 €	46,833.81 €	72,883.56 €
None	49,275.00 €	69,685.37 €	18,378.24 €	80,171.76 €
Long responders				
Not mentioned	60,764.84 €	29,538.65 €	47,668.13 €	73,861.54 €
Yes	98,389.07 €	16,110.32 €	91,246.16 €	105,531.99 €
NA	89,928.88 €	75,408.73 €	56,494.52 €	123,363.23 €
Dosage adjustment due to AEs active				
No	86,236.67 €	80,786.88 €	50,417.77 €	122,055.56 €
Not Relevant	83,546.67 €	76,674.75 €	49,550.99 €	117,542.34 €
Yes	72,825.08 €	47,833.71 €	51,616.80 €	94,033.36 €
Impact of treatment on autonomy				
No	50,990.95 €	35,392.51 €	35,298.79 €	86,289.74 €
Yes	112,407.66 €	55,550.03 €	87,778.15 €	137,037.16 €
Interval of treatment administration				
Daily	55,771.42 €	35,111.77 €	40,203.73 €	71,339.10 €
Weekly or less frequent	104,747.50 €	71,259.64 €	73,152.75 €	136,342.25 €
Variable treatment guideline				
No	76,789.96 €	54,409.58 €	52,666.11 €	100,913.81 €
Yes	74,671.70 €	55,243.71 €	50,178.01 €	99,165.38 €
Duration of treatment				
Fixed schedule	110,623.33 €	56,319.09 €	85,652.85 €	135,593.82 €
Other	89,918.15 €	66,683.08 €	60,352.53 €	119,483.78 €
Up to progression	56,666.91 €	36,126.07 €	40,649.51 €	72,684.32 €
Easy to use/mode & set of administration				
Injection	108,091.67 €	58.881.72 €	81.984.97 €	134.198.36 €
Intrathecal	189,430.00 €	- €	- €	- €
Oral	55,771.42 €	35,111.77 €	40,203.73 €	71,339.10 €
Combined chemotherapy				
With	108,549.34 €	59,703.00 €	82,078.51 €	135,020.17 €
Without	68,908.55 €	50,841.26 €	46,366.80 €	91,450.30 €
ESMO – MCBS setting curative/non-curative				
Curative	89,079.34 €	59,071.05 €	62,888.70 €	115,269.98 €
Non-Curative	73,235.22 €	53,406.60 €	49,556.06 €	96,914.38 €
ICER (> NICE threshold)				
No	71,603.22 €	14,199.42 €	64,542.02 €	78,664.43 €
Yes	82,283.45 €	59,142.59 €	52,872.53 €	111,694.36 €
ICER (NICE cancer fund)				
No	81,547.28 €	62,812.10 €	53,697.95 €	109,396.60 €
Yes	66,611.17 €	32,409.99 €	52,241.39 €	80,980.96 €
Recommendation by experts				
NA	63,000.00 €	69,753.90 €	28,821.22 €	97,178.78 €
Not Recommended	35,209.46 €	26,193.69 €	22,374.79 €	48,044.14 €
Recommended	88,901.03 €	49,442.76 €	64,674.53 €	113,127.54 €
Included in national/sub-national health plan				
Included	73,752.18 €	49,723.26 €	51,706.12 €	95,798.24 €
Not Included	86,753.01 €	75,706.10 €	53,186.81 €	120,319.22 €
Preferences of the population as a need?				
Identified	110,990.61 €	45,170.96 €	90,962.93 €	201,953.55 €
Not identified	68,366.05 €	52,976.65 €	44,877.51 €	91,854.58 €
Stakeholders' expression of interest & alignment				
Identified	98,322.90 €	48,297.69 €	76,908.91 €	119,736.90 €
Not identified	69,584.52 €	54,346.47 €	45,488.64 €	93,680.39 €
Impact of the intervention on environment – packaging, production				
NA	108,149.44 €	57,269.58 €	80,546.39 €	135,752.50 €
No	77,689.18 €	54,898.41 €	51,228.99 €	104,149.37 €
Yes	46,191.30 €	37,980.02 €	27,885.52 €	64,497.09 €
Healthcare services delivery change				
No	69,930.29 €	48,388.88 €	48,475.86 €	91,384.71 €
Yes	86,940.90 €	63,093.87 €	58,966.65 €	114,915.16 €
Societal acceptability of the decisions				
Identified	102,425.00 €	98,393.91 €	58,799.58 €	146,050.42 €
Not identified	73,485.06 €	50,562.17 €	51,067.05 €	95,903.07 €
All products				
Yearly price	76,115.97 €	53,353.38 €	52,460.40 €	99,771.53 €

SD, standard deviation; 95% CI, 95% Confidence interval; AEs, Adverse events; ESMO-MCBS, European Society of Medical Oncology – Magnitude of Clinical Benefit Score; ICER, Incremental Cost-effectiveness ratio; NICE, National Institute for Health and Care Excellence.

joint clinical assessments able to respond to all Member States' requirements must be produced at the EU level, ideally through consensus, and become part of multi-step national procedures. This new regulation enhances, in this way, the relevance of multiple domains (clinical, social, or economic) of assessment in the process of decision-making by national price and reimbursement organisms, EVIDEM being a solid starting point. From this perspective, further research is needed to standardize measures and determine the socially acceptable weights among EVIDEM dimensions, as well as its translation into economic values by dimension. So far, very limited experiences (48) have been tested with this broader approach aimed at more transparent and fair pricing but there is still a lack of solutions to tackle additional

TABLE 4 Univariate analysis of the association between listed prices of oncology drugs and the dimensions of MCDA and subitems within each dimension.

Dimensions and individual items	N	Correlation estimate	Lower 95% confidence limit	Upper 95% confidence limit	Value of *p* for H0: Rho = 0
1. Disease severity	**22**	**−00,29**	**−00,63**	**0,15**	**0,18**
Speed tumor growth	20	−0.26	−0.61	0.18	0.23
% Metastasized	22	−0.23	−0.60	0.21	0.29
Expected survival 5-years	22	−0.37	−0.68	0.06	0.08
Overall Survival	20	0.09	−0.34	0.49	0.68
Physical function and general health (SF36 - EQ5D - EORTC QLQ-C30)	12	−0.11	−0.50	0.33	0.63
2. Size of affected population	**22**	**0,17**	**−0,27**	**0,55**	**0,44**
Prevalence	22	0.23	−0.21	0.59	0.30
Incidence	22	0.16	−0.28	0.54	0.47
3. Unmet needs	**22**	**0,05**	**−0,38**	**0,46**	**0,81**
Treatment options	22	−0.07	−0.48	0.36	0.74
Type of standard of care	22	0.05	−0.38	0.46	0.81
4. Comparative effectiveness	**22**	**0,15**	**−0,29**	**0,54**	**0,50**
Progression-Free Survival observed	22	−0.14	−0.53	0.30	0.53
Progression-Free Survival difference compared to control	18	0.14	−0.30	0.53	0.52
Objective Response Rate (RECIST/MRD) observed	19	−0.35	−0.67	0.09	0.10
Objective Response Rate (RECIST/MRD) difference compared to control	14	0.20	−0.24	0.57	0.37
Complete response (RECIST/MRD) observed	20	−0.01	−0.43	0.41	0.96
Complete response (RECIST/MRD) difference compared to control	15	0.38	−0.04	0.69	0.07
Partial response (RECIST /MRD) observed	18	−0.15	−0.54	0.29	0.49
Partial response (RECIST /MRD) difference compared to control	13	0.27	−0.17	0.62	0.22
Long responders (Yes/no)	11	0.17	−0.27	0.55	0.44
Overall Survival observed	15	0.21	−0.23	0.58	0.33
Overall Survival difference compared to control	12	0.29	−0.15	0.63	0.18
5. Comparative safety/tolerability	**22**	**−0,13**	**−0,53**	**0,30**	**0,55**
Any Adverse Events observed	22	−0.18	−0.56	0.26	0.42
Any Adverse Events difference compared to control	16	0.04	−0.39	0.45	0.87
Non-Fatal Serious Adverse Events (>3) observed	22	0.15	−0.29	0.54	0.50
Non-Fatal Serious Adverse Events (>3) difference compared to control	16	−0.02	−0.44	0.40	0.91
Fatal Adverse Events (Grade 5 AEs) observed	21	−0.06	−0.47	0.37	0.78

(Continued)

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TABLE 4 (Continued)

Dimensions and individual items	N	Correlation estimate	Lower 95% confidence limit	Upper 95% confidence limit	Value of *p* for H0: Rho = 0
Fatal Adverse Events (Grade 5 AEs) difference compared to control	16	0.08	−0.35	0.48	0.72
Dosage adjustment due to adverse effects	22	0.06	−0.37	0.47	0.78
Treatment discontinuation (due to AEs) active	22	−0.25	−0.61	0.19	0.25
Treatment discontinuation (due to AEs) difference compared to control	17	−0.07	−0.48	0.35	0.74
Median duration of treatment	22	−0.49	−0.75	−0.09	**0.01**
Extent of exposure: Other indications, number of indications	22	−0.22	−0.58	0.22	0.31
6. Comparative patient-perceived health/PRO	**22**	**−0,14**	**−0,53**	**0,30**	**0,54**
HRQoL	14	0.37	−0.06	0.68	0.08
Impact on Autonomy	22	−0.45	−0.73	−0.04	**0.03**
Frequency of treatment (administered how often)	22	0.40	−0.03	0.70	0.06
Variable treatment schedule	22	0.02	−0.40	0.44	0.92
Time of treatment	22	0.41	−0.01	0.71	0.05
Easy to Use/Mode & Set of Administration	22	−0.48	−0.75	−0.08	**0.02**
Combined chemotherapy	22	−0.27	−0.62	0.17	0.21
7.a. Magnitude of preventive benefit	**18**	**0.16**	**−0.28**	**0.55**	**0.47**
Magnitude of preventive benefit	18	0.16	−0.28	0.55	0.47
7.b. Magnitude of therapeutic benefit	**22**	**0.13**	**−0.31**	**0.52**	**0.57**
Magnitude of therapeutic benefit	22	0.13	−0.31	0.52	0.57
8. Comparative cost consequences – cost of intervention	**22**	**−0,03**	**−0,45**	**0,39**	**0,87**
Incremental Cost-effectiveness ratio (ICER) over NICE threshold (yes/no)	21	−0.09	−0.49	0.34	0.69
ICER: NICE assigns cancer fund (yes/no)	22	−0.01	−0.43	0.41	0.95
ICER: NICE value (€ or pounds – with 95% CI)	18	−0.08	−0.49	0.35	0.70
11. Quality of evidence:	**22**	**−0,02**	**−0,44**	**0,40**	**0,91**
JADAD/ESMO assessment of quality (from 1 to 5, where 5 is the maximum)	22	−0.02	−0.44	0.40	0.91
12. Expert consensus/clinical practice guidelines	**17**	**0,56**	**0,17**	**0,79**	**0,00**
Availability of guidance for use and recommendation in guidance/by experts	17	0.56	0.17	0.79	*0.00*
13. Contextual criteria	**22**	**0,03**	**−0,40**	**0,44**	**0,90**
Mandate and scope of the healthcare system	22	−0.05	−0.46	0.38	0.81
Population priorities and access	22	0.35	−0.09	0.67	0.11
Common goal and specific interests	22	0.26	−0.18	0.61	0.24

(Continued)

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TABLE 4 (Continued)

Dimensions and individual items	N	Correlation estimate	Lower 95% confidence limit	Upper 95% confidence limit	Value of *p* for H0: Rho = 0
Environmental impact	19	0.01	−0.41	0.43	0.97
System capacity and appropriate use of intervention	22	−0.17	−0.55	0.26	0.43
Political/historical/cultural context	22	0.05	−0.38	0.46	0.83

Dimension 7 was analyzed separately for preventive and therapeutic benefits since these used different scoring. Dimensions 9 to 12 had a single item each, so the estimate for the dimension is the same as that of the item. Due to lack of data, dimensions number 9 "comparative cost consequences – other medical costs" and the corresponding item "Cost treatment (procedures and tests-physician visits-hospitalizations) / Year" and number 10 "comparative cost consequences –non-medical costs" and the corresponding item "Cost/Year" were not analyzed for correlation.

limitations, such as a potential disincentive effect on R&D efficiency discouraging future disruptive innovation.

5. Conclusion

Our exercise shows that, regardless of the paucity of explicative criteria on the decisions, the use of a standardized multidimensional framework allowed us to identify that the listed prices of new cancer products with a single first reimbursed indication in Spain are related to the type of standard of care, references to long-lasting responses, the convenience of use of the drug, and its impact on patient's autonomy, as well as contextual indicators such as the existence of previous clinical consensus. While individual items are relatively explanatory, grouping by the synthetic MCDA-EVIDEM dimensions does not improve explicative value or information.

Based on our results and the lack of detailed information on how Spanish healthcare authorities define price and reimbursement conditions of new onco-hematologic drugs, we propose that the implementation of MCDA-EVIDEM methodologies may help to capture and report additional factors generally not included in consolidated assessment frameworks, such as the European Network for Health Technology Assessment (EunetHTA) core model. It may be opportune to consider this in the upcoming revision of the Spanish regulation for health technology assessments and pricing and reimbursement procedures (49).

Data availability statement

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found in the article/Supplementary material.

Author contributions

DE: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. FT: Conceptualization, Data curation, Investigation, Methodology, Validation, Writing – review & editing. RV: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Validation, Writing – review & editing. GP: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Validation, Writing – review & editing. MO: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Validation, Writing – review & editing. DG: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Writing – review & editing. DV: Conceptualization, Data curation, Investigation, Methodology, Writing – review & editing. TP: Conceptualization, Data curation, Formal analysis, Investigation, Methodology. JT: Conceptualization, Supervision, Validation, Writing – review & editing. CP: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Supervision, Validation, Writing – review & editing.

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Conflict of interest

DE and MO are full-time employees of Sanofi. JT reports personal financial interest in the form of scientific consultancy roles for Array Biopharma, AstraZeneca, Bayer, Boehringer Ingelheim, Chugai, Daiichi Sankyo, F. Hoffmann-La Roche Ltd., Genentech Inc., HalioDX SAS, Hutchison MediPharma International, Ikena Oncology, Inspirna Inc., IQVIA, Lilly, Menarini, Merck Serono, Merus, MSD, Mirati, Neophore, Novartis, Ona Therapeutics, Orion Biotechnology, Peptomyc, Pfizer, Pierre Fabre, Samsung Bioepis, Sanofi, Scandion Oncology, Scorpion Therapeutics, Seattle Genetics, Servier, Sotio Biotech, Taiho, Tessa Therapeutics, TheraMyc, and Tolremo Therapeutics. Stocks: Oniria Therapeutics and educational collaboration with Imedex/HMP, Medscape Education, MJH Life Sciences, PeerView Institute for Medical Education, and Physicians Education Resource (PER).

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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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1265323/full#supplementary-material

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Perceptions of Chinese hospital leaders on joint commission international accreditation: a qualitative study

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Background: Joint Commission International (JCI) accreditation plays a significant role in improving the quality of care and patient safety worldwide. Hospital leadership is critical in making international accreditation happen with successful implementation. Little is known about how Chinese hospital leaders experienced and perceived the impact of JCI accreditation. This paper is the first study to explore the perceptions of hospital leaders toward JCI accreditation in China.

Methods: Qualitative semi-structured interviews were used to explore the perceptions of the chief operating officers, the chief medical officers, and the chief quality officers in five JCI-accredited hospitals in China. Thematic analysis was used to analyze the interview transcripts and identify the main themes.

Results: Fifteen hospital leaders participated in the interviews. Three themes emerged from the analysis, namely the motivations, challenges, and benefits related to pursuing and implementing JCI accreditation. The qualitative study found that eight factors influenced hospital leadership to pursue JCI accreditation, five challenges were identified with implementing JCI standards, and eight benefits emerged from the leadership perspective.

Conclusion: Pursuing JCI accreditation is a discretionary decision by the hospital leadership. Participants were motivated by prevalent perceptions that JCI requirements would be used as a management tool to improve the quality of care and patient safety in their hospitals. These same organizational leaders identified challenges associated with implementing and sustaining JCI accreditation. The significant challenges were a clear understanding of the foreign accreditation standards, making staff actively participate in JCI processes, and changing staff behaviors accordingly. The top 5 perceived benefits to JCI accreditation from the leaders' perspective were improved leadership and hospital safety, improvements in the care processes, and the quality of care and the learning culture improved. Other perceived benefits include enhanced reputation, better cost containment, and a sense of pride in the staff in JCI-accredited hospitals.

KEYWORDS

joint commission international, accreditation, leadership, Chinese hospital, qualitative study

Introduction

A healthcare accreditation program may serve as an external means to assess or improve the quality of care by evaluating the performance of a healthcare service organization against a set of standards. Accredited healthcare service organizations are renowned for high-quality care and patient safety (1). The Joint Commission formed JCI to provide international clients with education and consulting services in 1994. JCI published its first international quality standards for hospitals in 2000. Our study focuses on JCI accreditation, which extends the Joint Commission's mission and standards worldwide by helping healthcare services organizations outside the United States improve the quality of care and patient safety. Pursuing international accreditation is voluntary for any healthcare services organization in China. As of December 2022, forty-six Chinese healthcare services organizations accredited by JCI make China rank fifth on the list of countries with the highest number of JCI-accredited healthcare organizations and account for 5% of the total 946 JCI-accredited organizations worldwide. In China, forty-four private hospitals, one public hospital, and one homecare organization achieved and maintained JCI accreditation (2). Our study is the first qualitative study to explore Chinese hospital leaders' perceptions toward JCI accreditation.

We used a few electronic databases, such as PubMed, Embase, and Cochrane Library, to review the literature about the perceptions of JCI accreditation. Keywords used in different combinations include "Joint Commission International," "perception," and "JCI accreditation." The previous studies were conducted in certain countries, such as Saudi Arabia, Israel, Singapore, South Korea, Belgium, the United Arab Emirates, Panama, Lebanon, and Turkey. Research methods included the in-depth qualitative interview or a cross-sectional survey to explore the attitudes toward JCI accreditation from administrative staff, physicians, nurses, and other health professionals. JCI accreditation is a system approach that evaluates the capability of the entire healthcare organization to produce good results (3). Although JCI aims to extend the Joint Commission's standards worldwide, there were complaints that JCI standards are less stringent than those of the Joint Commission in the United States (4). Previous studies revealed a need for more agreement about the value of JCI accreditation. Most nurses generally had positive attitudes, but most physicians perceived fewer benefits of participating in JCI processes than nurses (5). However, the characteristics of JCI were insufficient in attracting and retaining nurses (6). Compared with healthcare professionals, the administrative staff was more satisfied with implementing JCI standards (7). Evidence indicated perceived benefits of pursuing and implementing JCI accreditation, such as improving the quality of training and education (8), building up quality improvement and patient safety culture (9), reducing variation in medical care (10), valuing the organizational change (5), and enhance hospital branding (11). However, there was no consensus among hospital managers about whether or not JCI accreditation had an enduring impact on the improvement (9). Side effects of implementing JCI standards were perceived, such as the preparation of JCI distracted healthcare workers from daily clinical work (8), requiring substantial monetary resources (10), and being time-consuming (11).

Compared with the local hospital accreditation standards and procedures, hospital professionals are less familiar with JCI accreditation, and international standards. Pursuing JCI accreditation is a leadership decision, but few studies have explored senior leaders' perceptions about JCI accreditation. The JCI accreditation standards have stricter requirements than the local accreditation standards. While JCI accreditation is unfamiliar to Chinese leaders and the standards more rigorous, Chinese leaders' perceptions of the value of JCI accreditation would be consistent with study findings in other countries. Besides, our study may explore different motivations, challenges, and perceived benefits from Chinese hospital leaders' perspectives.

Materials and methods

Design/methodology

Our study used a semi-structured interview approach to understand the perceptions of hospital leadership in JCI-accredited hospitals in China.

Ethical approval

Prior to starting the study, ethical permission was obtained by the Institutional Review Board of the Johns Hopkins Bloomberg School of Public Health.

Selecting participants

We applied the purposive sampling technique. Given a list of 40 JCI-accredited private hospitals in China, OBGYN hospitals accounted for 27.5% (11/40), the largest specialty hospital among accredited private specialty hospitals. The eligible participant would be the decision-maker in pursuing JCI accreditation and the leader in implementing JCI standards. Therefore, the leaders who hold any of the 3-key roles: Chief Operating Officer (COO), Chief Medical Officer (CMO), and Chief Quality Officer (CQO) were our targeted interviewees. After frequent contact, 15 leaders from five private OBGYN hospitals agreed to participate in in-depth interviews. The participants were given information that described the purpose of the study. They were asked to give oral informed consent to participate in the study before commencing the interviews. Table 1 lists the invited leaders and their positions. We labeled the interviewees by the hospitals and positions. For example, the COO at Hospital A is coded as COO1; the CMO at Hospital B is labeled as CMO2, and so on.

Data collection

An experienced qualitative researcher interviewed the participating executives at the hospital sites where they work, with two assistants during the process. The interviewer started by introducing themselves to the interviewee and asked permission to audio record the interview. The semi-structured questions (Table 2) were designed to elicit the participant's perceived motivations, the challenges, and the benefits of JCI accreditation. The interviewer reordered the semi-structured questions depending on the participant's flow of thought. The research assistants were responsible for taking notes about the conversation. The interview took 60 min on average.

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TABLE 1 Participant ($N = 15$) characteristics.

Hospital setting	Hospital A	Hospital B	Hospital C	Hospital D	Hospital E
- Number of participants	3	3	3	3	3
Position					
- Chief Operating Officer (COO)	1	1	1	1	1
- Chief Medical Officer (CMO)	1	1	1	1	1
- Chief Quality Officer (CQO)	1	1	1	1	1
Gender					
- Male	1	2	1	1	1
- Female	2	1	2	2	2
Highest level of education					
- Bachelor	1	1	1	1	2
- Master	1	1	0	1	0
- Doctorate	1	1	2	1	1

Data analysis

The verbal data of the audio recording were transcribed into the written text of transcripts. The notes taken from the interview can be used as a second opinion on the accuracy of the recording. We followed Braun and Clarke's six-phase process (12) and utilized MAXQDA 2022 for thematic analysis. Several stages (phases) are performed to do the analysis. First, we started the study by creating a new project on MAXQDA and importing the documents of interview transcripts into the new project. We read the entire data set a few times and identified semantic themes. Second, after understanding the data set, we used MAXQDA to code the data by tagging and naming text selections according to the features of the data. Then, we assigned codes to the data segments. Two-round coding was used to enhance the accuracy and coherence of coding. Two research assistants with training performed initial coding. An experienced qualitative researcher double-checked the codes. If inconsistency appears, a discussion will be used to reach an agreement. Third, we collated the relevant coded data extracts within identified themes. Within each theme, five to eight sub-themes were identified. Fourth, we re-read the entire data set to code any additional data within themes missed in earlier coding stages. Fifth, we finalized the themes and sub-themes by further refining those themes. Finally, with a set of fully worked-out themes, we completed the analysis, including suitable data extracts to demonstrate the prevalent themes. Visualizing the results is one of MAXQDA's strengths, and Code Matrix Browser showed the results in Figures 1–3.

TABLE 2 Semi-structured interview questions.

1	Is your organization JCI accredited?
2	Why is JCI accreditation important to your organization?
3	What are the motivations for your organization to pursue the JCI accreditation?
4	What are the challenges during the journey to JCI accreditation?
5	Can you describe the process for gaining buy-in to pursue JCI accreditation?
6	Has JCI accreditation improved your organization? 1) If so, please elaborate on examples 2) 2) If not, what was expected?
7	Does your organization support JCI accreditation? Why or why not?
8	What are the benefits of the JCI accreditation program?
9	What are your attitudes toward the JCI accreditation?
10	What are your observations post-JCI's survey in your organization?
11	What do you think about the difference between JCI and local standards?
12	What do you think are your colleague's opinions on the JCI program?

Results

Our thematic analysis generated three main themes to describe Chinese hospital leadership's perceptions of JCI accreditation. These are motivations for pursuing JCI accreditation, the challenges of implementing JCI standards, and the perceived benefits of JCI accreditation.

Theme 1: motivations for pursuing JCI accreditation

The participants identified eight sub-themes related to perceived motivators to pursue accreditation. Ten out of fifteen participants expected JCI accreditation to improve the safety culture. Eight out of

fifteen participants believed that JCI accreditation improved the quality of care and served as a management tool to improve performance. Figure 1 links sub-themes to types of professions. For example, chief medical officers expressed that JCI is motivated by improving hospital safety culture and improving the quality of care, in addition to improving hospital safety culture. In contrast, Chief operation officers recognized the motivation for JCI accreditation as a management tool.

Sample quotes from some participants in italics are as follows.

Use accreditation as a management tool

> "I can use JCI standards to manage physicians' behavior. Our physicians had different working experiences in other hospitals before they joined our hospital. Therefore, they had different behaviors or even mindsets in the process of medical care. I think JCI standards created a common language of care and patient safety among our physicians." (CQO1)

> "Our (Chinese) local hospital accreditation is a rigorous assessment, but it focuses on the medical outcomes. JCI accreditation evaluates the process of care but also assesses the hospital's governance. We learned effective leadership in the JCI program." (COO2)

Theme 2: challenges of implementing JCI standards

Even though leadership decided to pursue the accreditation, implementing JCI standards into routine work inevitably encountered challenges. Among the six challenges of implementing JCI standards, 11 out of 15 participants (73%) expressed difficulty understanding JCI standards precisely. This appeared to be the common issue of transforming the English-based criteria to another language and medical care system. Figure 2 indicates the main concerns about implementing JCI standards. Making staff actively participate in the accreditation process and changing staff mindset or behavior is ranked as the second challenge to implementing JCI accreditation.

Sample quotes from some participants in italics are as follows.

Understand JCI standards precisely

> "We knew little about JCI standards, which is a foreign accreditation. Thus, we engaged a consulting firm to help us prepare JCI examination. During the preparation of JCI accreditation, there were debates between the external consultants and our health care professionals about how and what to do to meet JCI requirements/standards." (CMO1)

> "We had to learn JCI standards on our own, but the Chinese version of JCI standards on our hands was difficult to understand. Our concern was the misunderstanding of JCI requirements" (CMO2)

> "When some physicians complained that some of JCI requirements could conflict with local practices, we did not know who was able to make the judgment." (COO1)

Change staff mindset or behavior

> "Some physicians argued that local accreditation has the same level of requirements regarding improving the quality of care and patient safety, and therefore it was not necessary to pursue JCI accreditation" (COO3)

> "Some staff got back to their old routines rather than strictly following JCI requirements in their daily works after the JCI survey (every three years) was passed." (CMO3)

Unfavorable perceptions regarding JCI accreditation

One unfavorable perception is the relatively higher standard or criteria in physical and environmental safety policy, as compared with the minimum requirement required by China's national or local requirements. Any infrastructure changes not only mean more money investment but also affect patients' volume and revenue during the construction period. Some participants in our study recalled that at

Code System	CQO	CMO	COO	Total
Theme 1: Motivations for pursuing JCI Accreditation				
Improve hospital safety culture	2	4	4	10
Take accreditation as a management tool	2	1	5	8
Improve the quality of care	1	4	3	8
Build a shared value and mission	2		3	5
Gain a distinguished reputation		2	2	4
Standardize the process of care	2		2	4
Obtain the latest international standards	1	1	1	3
Develop international medical services	1			1

FIGURE 1
Code matrix browser within theme one.

Code System	CQO	CMO	COO	Total
Theme 2: Challenges of implementing JCI standards				
Understand JCI standards precisely	4	4	3	11
Make satff actively participate in the accreditation process	2	2	3	7
Change staff mindset or behavior	2	1	4	7
Have a sufficiently qualified Workforce	3		2	5
Make significant investments	1	1	3	5
Prepare JCI examination in Covid-19 pandemic	1			1

FIGURE 2
Perceived challenges of implenting JCI standards.

the time of the preparation for JCI accreditation, they were struggling to decide to renovate the hospital's infrastructure and facilities to meet JCI requirements.

Another unfavorable perception was related to staff resistance to participating in JCI. One of the reasons behind the resistance could be the excessive documentation to meet JCI requirements. Both clinicians and other paramedical professionals complained that they have to spend more time on documentation, thereby taking some time from their patient care or patient-related work. In general, during the preparation, they need to work overtime to meet clinical work as well as the JCI documentation. Hospital executives encountered resistance from those staff who argued that some documentation was too excessive and unnecessary.

The third unfavorable perception happened during the JCI on-site examination. Few staff in hospitals can speak English fluently; however, JCI examiners are required to use English to interview staff. The hospital was responsible for having English translators during the on-site survey and interviews. The translators might not translate precisely both the questions or answers, which results in possible miscommunication or misunderstanding.

Theme 3: perceived benefits due to JCI accreditation

Ten out of fifteen participants indicated that JCI accreditation improved hospital leadership management and safety culture (Figure 3). In general, CMOs and COOs perceived JCI benefits more than CQOs. Followed by leadership and safety culture, improved care processes, and quality of care, promoting a learning culture were recognized by the participating hospital leaders.

Sample quotes from participants in italics are as follows.

Improved leadership and management

> "The chapter on GLD (Governance, Leadership, and Direction) in JCI Standards gave guidance on how to achieve effective leadership. Also, we can require teams to comply with a specific standard in the process of care by emphasizing that we worked in a JCI-accredited hospital." (COO2)

> "JCI standards helped optimize the existing processes. These effective processes can reduce waste and redundancies. I could not achieve the effective process alone. I have to more actively communicate with peers in different departments to find out what caused wastes, and then worked out solutions." (CMO3)

Improved safety culture

> "During the JCI program, we improved the mechanism for reporting an adverse event. We encourage our staff to identify potential risks and work out strategies and methods to prevent adverse events." (CQO2)

Discussion

The interrelated themes of JCI accreditation focused on the motivations for the change, challenges in the journey to the accreditation, and perceived benefits. The results indicated that Chinese hospital leaders were motivated by different goals when pursuing JCI accreditation. The prevalent motivations include using accreditation as a management tool to improve organizational performance, safety, and quality of care. However, the same leaders experienced difficulties in implementing the accreditation. The significant challenging tasks for the leaders are understanding the JCI standards precisely, making staff actively participate in the accreditation process, and changing their behaviors according to accreditation requirements.

There were a few relevant specific unfavored perceptions from leaders' perspectives. Stricter JCI criteria in the hospital physical safety environment require the hospital to spend money to upgrade the hospital's infrastructure and facilities and decrease the patient volume during the construction period. However, the investment would ultimately be helpful for patient safety, therefore differentiating the accredited hospital from the non-accredited hospital. Another study also found that resistance from medical staff was a significant issue in implementing JCI standards (5). This negative perception might be because team managers did not correctly understand the intention of individual JCI standards, and thereby, they could not know how to implement the JCI requirements effectively. Tarieh et al. (13) found that if hospital leaders ensure staff involvement with management decisions, staff motivation will increase. Then, staff resistance will be minimized, and staff productivity will increase (13). The increased unnecessary workloads lowered the staff's enthusiasm for JCI-related

Code System	CQO	CMO	COO	Total
Theme 3: Perceived benefits of JCI Accreditation				
Improved leadership and management	2	5	3	10
Improved safety culture	2	4	4	10
Improved care processes	3	3	2	8
Improved quality of care	2	1	4	7
Promoted a culture of learning	2	3	2	7
Better reputation in the public	1	1	2	4
Better cost containment	1		2	3
Enhanced the sense of pride	1		1	2

FIGURE 3
Perceived benefits of JCI accreditation.

tasks. For example, some staff complained that excessive documentation due to JCI requirements was unnecessary. Leaders should consider how the accreditation process may improve the documentation and decrease the staff's workload. One of JCI accreditation's characteristics could be its international, as its English standards are translated and applied in different countries and cultures worldwide. As few Chinese hospital staff have qualified English to understand foreign standards, translation is inevitable. Unqualified translation words could cause misunderstanding unless JCI reviewers can use the local language to conduct on-site surveys.

On the other hand, participants in our study perceived the benefits of JCI accreditation. The improved management leadership and safety culture were the two most benefits perceived by hospital leaders. De Meester et al. (14) indicated that JCI program improved the communication between the nurses and physicians, resulting in a lower number of unexpected deaths. Novarro-Escudero et al. (15) found that Under JCI guidelines, almost all stroke core measures continuously improved over three years. However, the observed improvements in accredited hospitals may not necessarily be attributed to the accreditation (16), and benefits to patient safety should not be resulted from hospital accreditation only (17). Getting international accreditation motivated some participants in our study, and they felt a distinguished reputation when their hospitals achieved JCI accreditation. This result is consistent with the finding that although healthcare professionals felt stressed about achieving the accreditation, they were proud after they went through the accreditation process (18). However, one study suggested that achieving accreditation cannot guarantee that the accredited hospital provides high-quality care continuously (18).

Our study's implications are consistent with previous studies. First, patient safety and quality of care is the most prominent benefit of JCI accreditation as perceived by leaders. Therefore, from a macro level, in the context of upgrading a national healthcare system to achieve a good quality of care, the study findings may provide references to policymakers, as well as hospital management decision-makers, to consider JCI accreditation as a management tool. Good leadership and organizational culture facilitate JCI accreditation implementation (19). Top management is vital for successful implementation and effectiveness (20). Hospital leadership commitment and effective governance are essential to effectively implementing JCI accreditation (21). Financial support is needed to have qualified infrastructure, equipment, and staff education to comply with JCI requirements. Staff resistance was seen as a big challenge in implementing accreditation. One possible reason for resistance could be that there were no monetary or other incentives to encourage healthcare professionals or administrators to participate in the JCI program. Another study pointed out that the resources to achieve accreditation should be based on support from coworkers and managers (22). The findings in our study implied that leaders are responsible for leading staff across different departments to collaborate to achieve accreditation by allocating monetary and human resources in the accreditation program. Although the implementation of accreditation requires all staff participation in the hospital, its leaders should be accountable for driving the process (18). If the hospital leadership showed commitment to the process, the implementation of accreditation would be easier, especially when hospital leaders involved staff in the process (7). In addition to commitment in the process, another way to overcome the resistance issue is the education or dissemination of JCI's impact on medical care quality, as well as timesaving for workers' daily routine, which might reduce the resistance toward JCI accreditation. To address the challenge of language barriers, the hospital's top executives might implement a periodic review of the JCI accreditation standards or procedures to enhance the comprehension of the standard in a selected group of members.

Our survey participants are limited to the hospital's top management; their perception is not subjected to the opinion of the first-line managers or employees. In general, first-line healthcare workers are persons who carry the daily and prepare or execute the JCI policy and procedures. The future study is suggested to interview to solicit perceptions of physicians, nurses, and first-line managers. We will develop a research questionnaire based on the in-depth interview results and survey the same hospital workers to examine their perceptions on JCI accreditation.

Limitations

As the study was limited to a few hospitals in China, the results could not be relevant to other settings. However, the study aimed to understand Chinese hospital leaders' perceptions, thereby adding to the current literature. Another limitation is that the study only included private hospitals, and the selection bias made the findings in the study not generalizable to public hospitals. However, the public hospital only

accounts for 2% of JCI-accredited hospitals in China. Public hospitals in China are less pursuing the JCI accreditation than the private hospital's counterpart, as quality improvement strategy. Although hospital numbers are about equal in both sectors, the scale of public hospitals is larger than that of private hospitals. The complexity of management increase with the number of bed. The study only limited to private hospital leaders' perception; if a similar study is conducted at public hospitals, a comparison could be derived for the larger healthcare system. The third limitation is that the number of participants in our study is small; they could not represent the leadership in other private hospitals. However, the participants provided rich information about JCI accreditation initiatives and implementation, consistent with the literature. However, the participants who accepted the interviews might favor the JCI accreditation policy over those who refused to participate in the study (23).

Conclusion

The impact of JCI accreditation has been widely studied in different countries, but few qualitative studies have been conducted in Chinese hospitals. Our paper is the first study to explore the perceptions of leadership in JCI-accredited hospitals in China on motivation, challenges, and benefits of JCI accreditation. Despite its limitations, the study contributed to the current literature. Our study indicated that the leaders in Chinese private hospitals value JCI accreditation as a management tool based on patient-centeredness to better treat individual patients and their family members. The same organization leaders encountered one of the challenges in leading the JCI program: staff resistance. One of the reasons behind the opposition could be the issue of interpreting JCI standards. Not properly understanding JCI requirements would cause unnecessary workload and waste resources. It lowered staff enthusiasm to participate in the accreditation. Also, our study added to the understanding of leaders' thoughts when they planned the accreditation and their experiences during the implementation of accreditation. These findings provide insight that dealing with those difficulties during the accreditation process may improve leadership and hospital culture.

Data availability statement

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

Ethics statement

Ethical permission was obtained from the Institutional Review Board (IRB) at the Johns Hopkins Bloomberg School of Public Health (reference number IRB00021496). The participants were given information that described the purpose of the study. They were asked to give oral informed consent to participate in the study before commencing the talk.

Author contributions

HZ: Conceptualization, Formal analysis, Writing – original draft. ST-H: Data curation, Writing – review & editing. MB: Supervision, Writing – review & editing. LE: Writing – review & editing. HC-C: Conceptualization, Methodology, Writing – review & editing.

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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.

Publisher's note

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Health care costs of cardiovascular disease in China: a machine learning-based cross-sectional study

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Background: Cardiovascular disease (CVD) causes substantial financial burden to patients with the condition, their households, and the healthcare system in China. Health care costs for treating patients with CVD vary significantly, but little is known about the factors associated with the cost variation. This study aims to identify and rank key determinants of health care costs in patients with CVD in China and to assess their effects on health care costs.

Methods: Data were from a survey of patients with CVD from 14 large tertiary grade-A general hospitals in S City, China, between 2018 and 2020. The survey included information on demographic characteristics, health conditions and comorbidities, medical service utilization, and health care costs. We used re-centered influence function regression to examine health care cost concentration, decomposing and estimating the effects of relevant factors on the distribution of costs. We also applied quantile regression forests—a machine learning approach—to identify the key factors for predicting the 10th (low), 50th (median), and 90th (high) quantiles of health care costs associated with CVD treatment.

Results: Our sample included 28,213 patients with CVD. The 10th, 50th and 90th quantiles of health care cost for patients with CVD were 6,103 CNY, 18,105 CNY, and 98,637 CNY, respectively. Patients with high health care costs were more likely to be older, male, and have a longer length of hospital stay, more comorbidities, more complex medical procedures, and emergency admissions. Higher health care costs were also associated with specific CVD types such as cardiomyopathy, heart failure, and stroke.

Conclusion: Machine learning methods are useful tools to identify determinants of health care costs for patients with CVD in China. Findings may help improve policymaking to alleviate the financial burden of CVD, particularly among patients with high health care costs.

KEYWORDS

health care costs, cardiovascular disease, quantile regression forest, machine learning, financial burden

Introduction

Cardiovascular disease (CVD) refers to a group of disorders of the heart and blood vessels, including coronary heart disease (CHD), cerebrovascular disease, peripheral arterial disease, rheumatic heart disease, congenital heart disease, deep vein thrombosis, and pulmonary embolism. CVD is the leading cause of death globally (1). The World Health Organization (WHO) has reported that an estimated 17.9 million people died from CVD in 2019, representing 32% of all global deaths (2). Over three quarters of CVD deaths take place in developing countries (2). In China, it was estimated that about 330 million patients suffer from CVD and two out of every five deaths were due to CVD (3).

Besides the burden of morbidity and mortality, CVD results in substantial financial burden to patients and their families in China (4, 5). There is emerging evidence that CVD and other noncommunicable diseases can lead to poverty due to the high health care cost of treating the disease and high out-of-pocket expenditure among those who are uninsured or underinsured (6). Previous studies have assessed the health care cost of treating one type of CVD such as hypertension (7–10) and CHD (11–13) and mostly focused on developed countries such as the United States (6, 14–16). No studies, to the best of our knowledge, have assessed the health care costs of treating all types of CVD in China. The current study aims to fill this research gap by assessing the health care costs in patients with all types of CVD in China.

In previous health care cost analyses of CVD, mean-based models (e.g., generalized linear models with certain link functions for specific distributions) are often used to identify factors associated with the mean health care costs of CVD (17–19). These models evaluate the relationship between covariates and the mean outcome, assuming a uniform relationship across different percentiles of the cost distribution (20). However, this assumption does not always hold because the determinants of high health care costs may be different from those of low costs, and the effects of the determinants may vary across different parts of the cost distribution. The current study, instead, uses a machine learning approach to identify key determinants of health care costs in patients with CVD in China. We hypothesize that the determinants of health care cost vary from patients with high costs to those with low costs. The results would help healthcare professionals and policymakers design targeted interventions that may alleviate the financial burden of patients with CVD, their households, and the health care system.

Methods

Data sources and study population

This study used a cross-sectional cohort design. We exacted data from a survey of patients with CVD from 14 tertiary grade-A general hospitals in S City between 2018 and 2020. The list of hospitals is described in Supplementary Table S1. The survey collected patient information on their demography (e.g., age, sex), health conditions and comorbidities, medical service utilization, and health care costs.

Our sample included 28,213 adult patients ≥18 years. Excluding 591 patients with missing values on key variables, the final sample included 27,622 patients (Supplementary Figure S1). The International Classification of Diseases, 10th revision (ICD-10) codes I00-I99 and Q20-Q28 were used to identify patients with CVD (21). Specific types of CVD included congenital heart disease (ICD-10 codes Q20-Q28), coronary artery disease (CAD) (ICD-10 codes I25), heart failure (ICD-10 codes I50), myocardial infarction (MI) (ICD-10 codes I21), cardiomyopathy (ICD-10 codes I42-I43), hypertensive disease (ICD-10 codes I10-I15), and stroke (ICD-10 codes I60, I61, I63, I64) (Supplementary Table S2) (22). The survey data are de-identified. An exemption from ethical review has been approved by the Institutional Review Boards of School of Public Health at Shanghai Jiao Tong University School of Medicine.

Outcomes

The outcomes of interest included total health care cost, which consists of insurance-covered cost and out-of-pocket cost. We also considered health care costs by treatment category, including the cost of comprehensive medical service, diagnosis, treatment, medication, and medical consumables.

Covariates

The covariates included age, sex, marital status, residency (living in S City, living outside of S City), employment status (retired, employed, unemployed), common diagnosis (CHD, CAD, heart failure, MI, cardiomyopathy, hypertensive disease and stroke), cardiogenic comorbidity (yes, no), non-cardiac comorbidity (yes, no), number of comorbidities (0, 1, 2, 3, 4, ≥5), number of medical procedures, level of operation (I, II, III, IV), length of hospital stay, payment method (Urban Employee Basic Medical Insurance (UEBMI), New Rural Cooperative Medical Scheme/Urban Resident Basic Medical Insurance (NRCMS/URBMI), full self-payment and others), admission type (emergency and outpatient), and proportion of out-of-pocket.

Statistical analysis

To describe the distribution of health care costs, we first calculated the means of different quantiles of annual health care costs by demographic characteristics and compared the differences. We also calculated the Gini coefficient, a measure of inequality, for health care costs, the proportion of high health care costs (top 10%) in total costs. A high Gini coefficient suggests inequality in health care costs. We examined the statistical differences between high- (top 10%) and low-cost patients (bottom 10%) by between-group difference tests. We used recentered influence functions (RIFs) to estimate the small changes in the distribution of independent variables on the distributional measure of interest such as Gini coefficient (23).

We used quantile regression forests (QRFs) to identify determinants of health care costs across patients with different quantiles of CVD costs. Quantile regression (QR) can estimate how specific quantiles or percentiles of the distribution of the outcome variable vary with covariates. It is robust against outliers and is more informative for a skewed distribution than mean-based regression (24). QRFs is an extension of QR and has been applied in health care

research for its prediction accuracy (25). QRFs utilizes the infrastructure of random forests and is a nonparametric model for conditional quantile estimation.

Specifically, we implemented a backward stepwise variable selection algorithm developed by Hu et al. (26), based on the variable importance scores generated by QRFs to determine the key factors for the 10th, 50th, and 90th quantiles, respectively, representing the low-, and high cost threshold of health care costs in patients with CVD (25). We computed the importance score of a QRFs for each covariate based on the "mean decrease in accuracy" (20). In a QRFs ensemble, each tree has an out-of-bag (OOB) sample that was left out from tree construction for assessing the predictive performance of the tree model (25). An iterative process was carried out for variable selection. Each time, we removed the least important variable and rebuilt a QRFs model with the remaining variables and recorded the out-of-bag average quantile loss (AQL) until no variable was left. AQL was used for the evaluation of model performance. Finally, we performed a weighted quantile regression (QR) to quantify the effects of each selected factor on different quantiles of health care costs in patients with CVD. Taking into account the variance across the specific CVD diseases, we performed subgroup analyses of CAD and stroke, the two diseases with the highest prevalence. As a sensitivity analysis, we also used GLM to estimate the effect of the key factors on the medical expenditures by the mean-based approach.

All statistical analyses were performed by R version 4.2.2. QRFs models were built using the "quantregForest" R package. A p value from two-sided test <0.05 was considered statistically significant.

Results

Population characteristics and distribution of health care costs

Out of 27,622 patients with CVD, 10,614 (38%) were female and 19,427 (70%) lived in S City. The mean age of the patients was 64.14 (14.09) years. The proportion of males among the top 10% of patients was higher than among the bottom 10% of patients (64% vs. 56%), and the same applied to the proportion of non-native (46% vs. 31%). Other population characteristics were summarized in Table 1.

Nearly 38% of the health care costs for treating CVD were generated by the top 10% of patients and 1% by the bottom 10%. The Gini coefficient of health care costs in patients with CVD was 0.56, that of out-of-pocket cost was 0.74, and that of medical insurance cost was 0.65, indicating a very high concentration. The Gini coefficient of health care costs for additional factors were showed in Supplementary Table S3. Among all types of CVD, the Gini coefficients of the costs exceeded 0.4 except for MI. The Gini coefficient of cardiomyopathy was the largest, reaching 0.68 in total cost, 0.82 in out-of-pocket, and 0.76 in medical insurance, which was much higher than the average (Figure 1).

The average health care cost was 41,282 CNY, while the 10th, 50th, and 90th of the costs were 6,103 CNY, 18,105 CNY, and 98,637 CNY, respectively. The average health care costs for each type of CVD were presented in Supplementary Table S4. Among them, myocardial infarction (MI) had the highest average annual per-person cost (56,118 CNY). Hypertensive disease had the highest cost (4,533 CNY) in the low-cost group, while cardiomyopathy was highest in the high-cost group. Coronary artery disease (CAD) accounted for the highest proportion (21.04%) of total costs (Supplementary Table S4). The bottom 10% of inpatients had a higher proportion of insurance coverage, accounting for 93.8%, while the top 10% had 72.3%. Among the specific categories of health care costs, the bottom 10% of patients had the highest proportion of diagnostic costs, reaching 62.8%, while the top 10% of patients had the highest proportion of medical consumables, reaching 62.9% (Supplementary Table S5).

Variable selection and rank of importance in different quantiles

Figures 2A,C,E showed the estimated out-of-bag AOL from every QRFs model built at each iteration in the backward stepwise algorithm for the 10th, 50th, and 90th quantiles of health care costs in patients with CVD. Number of operations, level of operation, inpatient length of stay, admission type and residence were selected as important factors for patients with both the 10th percentile, 50th percentile, and 90th percentile healthcare costs. Number of comorbidities was selected as an important factor only for patients with the 10th percentile cost, while sex, age, medical payment and proportion of self-payment were selected only for patients with the 90th percentiles costs. Figures 2B,D,F plotted the importance scores for the selected key factors for the 10th, 50th, and 90th quantiles of health care costs in patients with CVD. The variable selection of the identified key factors and the estimated effect of selected key factors for the 10th, 50th, and 90th percentile of health care costs with CAD and stroke were presented in Supplementary Figures S2, S3 and Supplementary Tables S7, S8.

Estimated effect of selected key factors

Table 2 presented the estimated effects of the identified key determinants on total health care costs in patients with CVD. Compared to females, males were associated with higher costs among the high-cost groups (1,615 CNY, 95%CI: 348 CNY, 2,883 CNY). The cost would increase by 162 CNY (95% CI: 117 CNY, 207 CNY) per year of age among the high-cost group, but age was not significantly associated with health care costs in the low-cost group. The number of comorbidities was significantly associated with health care costs among the low-cost group. Patients with one, two, three, four or more comorbidities would have additional costs of 2,026 CNY (95% CI: 1,582 CNY, 2,471 CNY), 2,232 CNY (95% CI: 1,842 CNY, 2,622 CNY), 2,564 CNY (95% CI: 2,171 CNY, 2,957 CNY), and 2,635 CNY (95% CI: 2,267 CNY, 3,002 CNY), respectively, compared to those with no comorbidities. However, the number of comorbidities was not associated with costs among the high-cost groups. Payment methods were associated with costs in the high-cost groups, but not in the low-cost groups. Compared to UEBMI, NRCMS/URBMI was associated with higher costs (5,545 CNY, 95% CI: 660 CNY, 10,430 CNY), while this association was opposite in fully self-payment (−5,673 CNY, 95% CI: −7,841 CNY, −3,505 CNY). For every 1% increase in the proportion of self-payment, the costs increased by 134 CNY (95% CI: 100 CNY, 167 CNY) among the high-cost groups. Results from the sensitivity analysis using mean-based approach (GLM) were presented in Supplementary Table S9.

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TABLE 1 Demographic, disease characteristics and medical service utilization of inpatients with CVDs (n = 27,622).

Characteristics	All inpatients	The bottom 10%	The bottom 10–50%	The top 10–50%	The top 10%
Demography characteristics					
Sex, n (%)					
Male	17,008 (62)	1,537 (56)	6,441 (58)	7,265 (66)	1,765 (64)
Female	10,614 (38)	1,225 (44)	4,608 (42)	3,784 (34)	997 (36)
Age, mean (SD)	64.14 (14.09)	63.04 (15.73)	64.50 (13.94)	64.25 (14.19)	63.40 (12.38)
Marriage, n (%)					
Unmarried	1,168 (4)	132 (5)	312 (5)	473 (4)	84 (3)
Married	23,369 (85)	2,425 (88)	2,425 (88)	9,298 (84)	2,337 (85)
Widowed or divorced	3,085 (11)	205 (7)	205 (7)	1,278 (12)	341 (12)
Residence, n (%)					
Non-native	8,195 (30)	845 (31)	2,565 (23)	3,506 (32)	1,279 (46)
Native	19,427 (70)	1,917 (69)	8,484 (77)	7,543 (68)	1,483 (54)
Occupation, n (%)					
Retired employed	10,385 (38)	975 (35)	4,460 (40)	4,064 (37)	886 (32)
Employed	16,100 (58)	1,688 (61)	6,201 (56)	6,471 (59)	1,740 (63)
Unemployed	1,137 (4)	99 (4)	388 (4)	514 (5)	136 (5)
Disease characteristics					
Common disease, n (%)					
Hypertensive disease	1,521 (6)	336 (12)	951 (9)	216 (2)	18 (1)
Coronary artery disease (CAD)	7,239 (26)	592 (21)	3,307 (30)	2,964 (27)	376 (4)
Cardiomyopathy	291 (1)	60 (2)	133 (1)	53 (0)	45 (2)
Heart failure	542 (2)	55 (2)	281 (4)	170 (2)	36 (1)
Stroke	3,177 (12)	100 (4)	1,588 (15)	1,068 (10)	321 (12)
Congenital heart disease (CHD)	771 (3)	79 (3)	224 (2)	383 (3)	85 (3)
Myocardial infarction (MI)	946 (3)	15 (1)	92 (1)	761 (7)	78 (3)
Comorbidity type					
Cardiogenic comorbidity, n (%)					
No	12,167 (44)	1,415 (51)	5,304 (48)	4,519 (41)	929 (34)
Yes	15,455 (56)	1,347 (49)	5,745 (52)	6,530 (59)	1,833 (66)
Non-cardiac comorbidity, n (%)					
No	19,313 (70)	1,884 (68)	7,335 (66)	7,906 (72)	2,188 (79)
Yes	8,309 (30)	878 (32)	3,714 (34)	3,143 (28)	574 (21)
No. of comorbidities, n (%)					
0	3,852 (14)	537 (19)	1,588 (14)	1,373 (12)	354 (13)
1	4,352 (16)	487 (18)	1,716 (16)	1,703 (15)	446 (16)
2	5,079 (18)	520 (19)	1,987 (18)	2,060 (19)	512 (19)
3	4,448 (16)	424 (15)	1,715 (16)	1,860 (17)	446 (16)
≥4	9,894 (36)	794 (29)	4,043 (37)	4,053 (37)	1,004 (36)
No. of medical procedures					
0	7,920 (29)	2,270 (82)	4,380 (40)	1,247 (11)	23 (1)
1	8,493 (31)	424 (15)	4,926 (45)	2,777 (25)	366 (13)
2	4,455 (16)	57 (2)	1,089 (10)	2,757 (25)	552 (20)
3	3,185 (12)	9 (0)	571 (5)	1,997 (18)	608 (22)

(Continued)

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TABLE 1 (Continued)

Characteristics	All inpatients	The bottom 10%	The bottom 10–50%	The top 10–50%	The top 10%
≥4	3,569 (13)	2 (0)	83 (1)	2,271 (21)	1,213 (44)
Level of medical procedure, n (%)					
I	3,119 (15)	517 (60)	2,157 (31)	421 (4)	24 (1)
II	4,167 (21)	123 (14)	2,223 (32)	1,644 (17)	177 (7)
III	6,003 (30)	123 (14)	2,381 (34)	2,943 (31)	556 (21)
IV	6,893 (34)	103 (12)	285 (4)	4,586 (48)	1,919 (72)
Medical service utilization					
Length of hospital stay, mean (SD)	7.73 (9.41)	3.69 (2.43)	5.81 (4.06)	8.32 (8.24)	17.09 (20.54)
Medical payment, n (%)					
UEBMI	16,071 (58)	1,551 (56)	6,983 (63)	6,284 (57)	1,253 (45)
NRCMS/URBMI	2,595 (9)	258 (9)	1,062 (10)	977 (9)	298 (11)
Full out-of-pocket	6,473 (23)	761 (28)	2,357 (21)	2,579 (23)	776 (28)
Others	2,483 (9)	192 (7)	647 (6)	1,209 (11)	435 (16)
Admission type, n (%)					
Emergency	6,649 (24)	365 (13)	2,381 (22)	3,012 (27)	891 (32)
Outpatient	20,973 (76)	2,397 (87)	8,668 (78)	8,037 (73)	1,871 (68)
Proportion of out-of-pocket, *n* (%)	33.06 (37.29)	30.71 (40.92)	29.16 (37.61)	35.27 (35.53)	42.22 (36.81)

Level of medical procedure: I: a variety of surgeries with low technical difficulty, simple surgical procedure and low risk; II: all kinds of surgery with average technical difficulty, uncomplicated surgical procedure and medium risk. III: all kinds of surgery with relatively high technical difficulty, complicated surgical process and high risk. IV: all kinds of surgery with high technical difficulty, complicated surgical process and high risk. The level of operation is the highest of all operations. UEBMI, urban employee basic medical insurance; NRCMS, new rural cooperative medical system; URBMI, urban resident basic medical insurance.

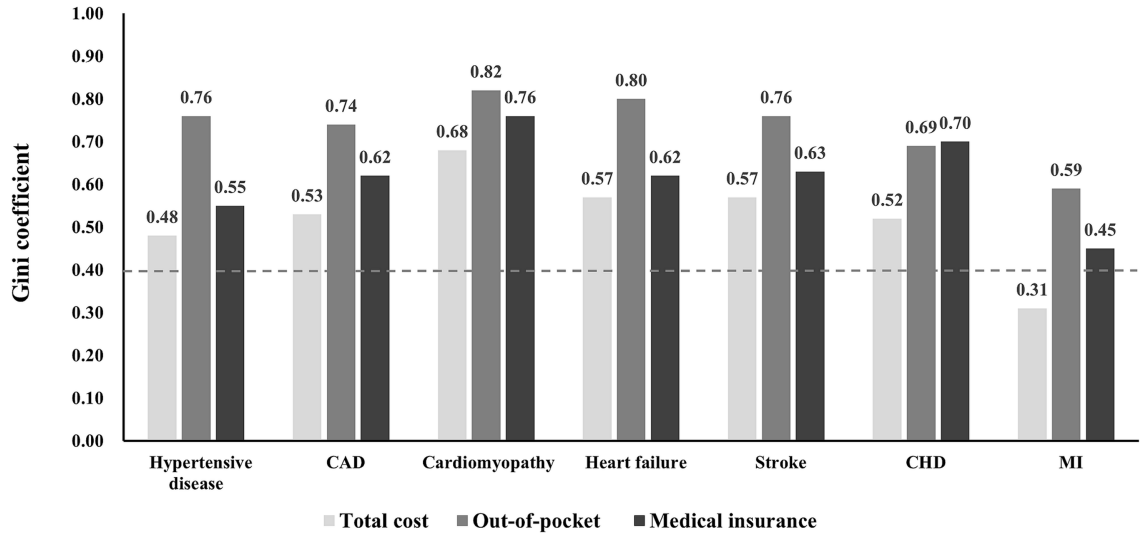

FIGURE 1
Gini coefficient of health care cost.

Discussion

The determinants of health care costs in patients with CVD in China are poorly known. Using data from a survey of patients with CVD from 14 large hospitals in China, we identified the determinants of health care costs in patients CVD and assessed their effects on the costs. The results showed that the health care costs in urban patients with CVD in China were highly concentrated in small groups of patients, similar to the empirical evidence (4, 27–29). The top 10% of inpatients accounted for approximately 38% of annual health care costs, and only 8% of the insured

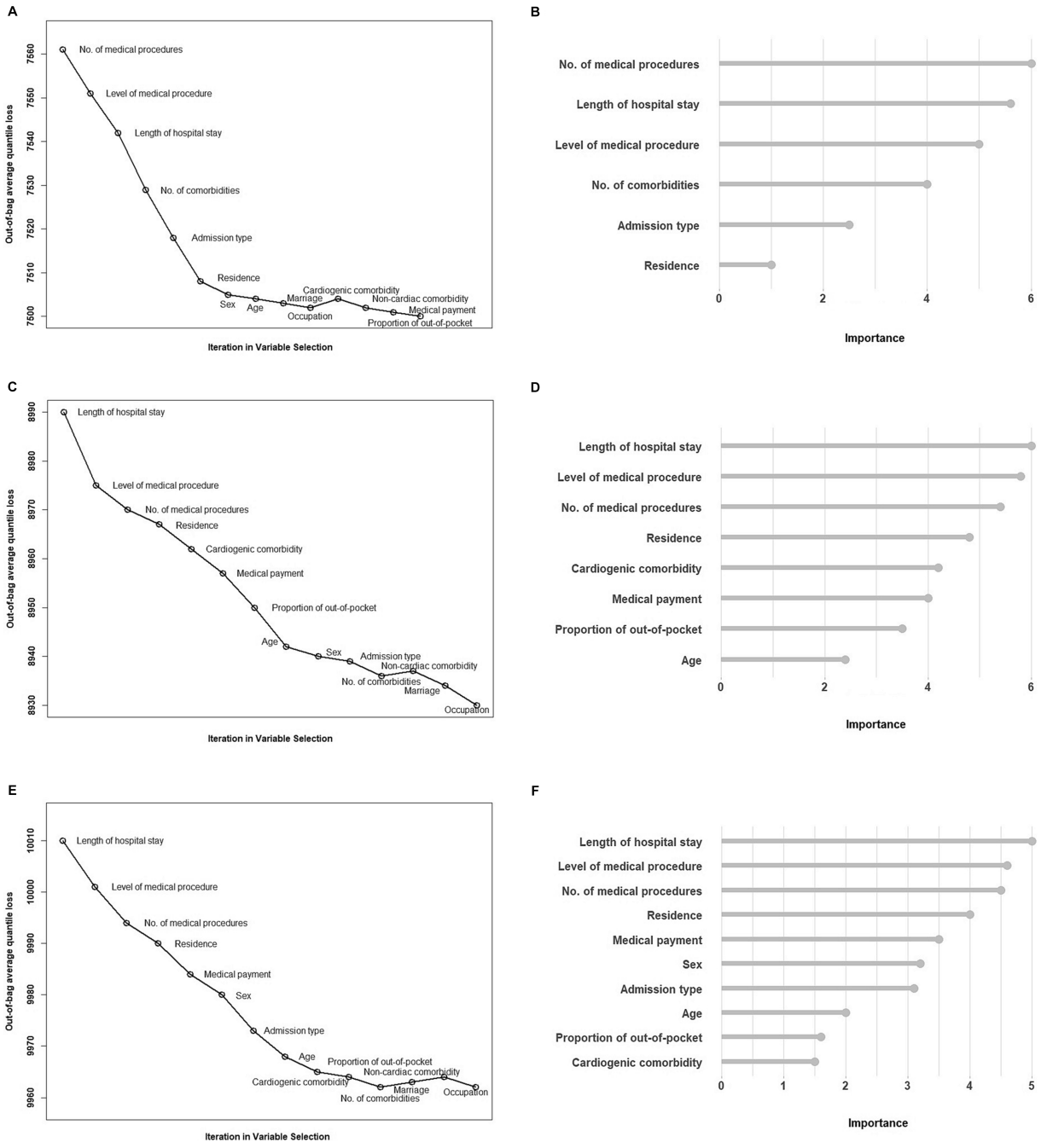

FIGURE 2
(A), **(C)** and **(E)** plotted the estimated out-of-bag AOL from every QRFs model built at each iteration in the backward stepwise algorithm for the 10th, 50th and 90th quantiles of health care costs in patients with CVD. **(B)**, **(D)** and **(F)** plotted the importance scores for the selected key factors for the 10th, 50th and 90th quantiles of health care costs in patients with CVD.

(UEBMI/NRCMS/URBMI) consumed close to 61% of annual inpatient medical insurance expenditures, which was generally consistent with the conclusions of some studies about other diseases in China (4, 30). Our study found that there was no difference in the concentration of total costs among age groups, but the concentration of out-of-pocket expenses and medical insurance expenses was completely opposite. The older adults have the highest concentration degree of out-of-pocket expenses and the lowest concentration degree of medical insurance expenses, although some evidence suggesting that concentration decreased with age (4, 31). Sex differences existed in the concentration of inpatient medical expenditures, with females having a greater concentration than males at all expense categories. Among the seven common diseases, patients with cardiomyopathy had the highest concentration and ones with MI had the lowest. Our results showed that high-cost inpatients were male and older, which explained the increase in the concentration of inpatient health care costs with population aging.

Employing the rigorous methodology to identify determinants for health care costs is important to informing the debate regarding how

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TABLE 2 Estimated effect of selected key factors on the medical expenditures using quantile regressions.

Characteristics	10th Quantile	50th Quantile	90th Quantile
Demography characteristics			
Sex			
Female vs. male	Not select	Not select	−1744.0 (−2976.8, −511.2)
Age	Not select	32.9 (19.1, 46.7)	138.7 (94.2, 183.2)
Marriage: ref. = Unmarried			
Married	Not select	Not select	Not select
Widowed or divorced	Not select	Not select	Not select
Residence			
Native vs. non-native	−487.5 (−722.5, −252.5)	−2660.8 (−3389.4, −1932.1)	−46629.7 (−6753.8, −2505.6)
Occupation: ref. = Retired			
Employed	Not select	Not select	Not select
Unemployed	Not select	Not select	Not select
Cardiogenic comorbidity			
Yes vs. No	Not select	1331.2 (765.5, 1897.0)	2955.8 (1507.5, 4404.1)
Non-cardiac comorbidity			
Yes vs. No	Not select	Not select	Not select
No. of comorbidities: ref. = 0			
1	2026.4 (1581.6, 2471.2)	Not select	Not select
2	2231.9 (1842.3, 2621.5)	Not select	Not select
3	2563.9 (2171.3, 2956.5)	Not select	Not select
≥4	2634.7 (2267.3, 3002.1)	Not select	Not select
No. of medical procedures: ref. = 0			
1	3556.0 (1899.7, 5212.2)	4118.0 (3604.4, 4631.5)	4301.3 (3249.6, 5352.9)
2	6793.5 (5127.7, 8459.4)	15314.7 (13727.0, 16902.4)	25434.5 (22903.8, 27965.2)
3	9264.9 (7590.0, 10939.8)	20719.0 (18832.8, 22605.2)	21383.0 (17899.2, 24866.8)
≥4	21279.9 (19660.8, 22899.0)	36196.5 (34394.6, 37998.5)	43420.3 (38433.7, 48406.8)
Level of medical procedure: ref. = I			
II	2605.9 (2320.3, 2891.5)	1228.1 (832.1, 1624.0)	31129.8 (28348.9, 33910.7)
III	2717.5 (2430.0, 3005.0)	6127.4 (5381.0, 6873.7)	39079.9 (36676.1, 41483.6)
IV	15668.0 (14480.0, 16855.9)	30406.2 (29288.0, 31524.3)	73859.6 (70215.4, 77503.8)
Medical service utilization			
Length of hospital stay	8288.0 (7516.5, 9059.5)	27299.2 (26155.8, 28442.6)	446134.6 (43871.2, 48398.0)
Medical payment: ref. = UEBMI			
NRCMS/URBMI	Not select	−36.8 (−739.1, 665.5)	5459.9 (1296.5, 9623.3)
Full out-of-pocket	Not select	−3333.9 (−3976.2, −2691.5)	−5721.2 (−7699.5, −3742.8)
Others	Not select	1324.3 (13.3, 2635.3)	918.7 (−1923.4, 3760.8)
Admission type			
Outpatient vs. Emergency	−907.6 (−1243.7, −571.5)	Not select	−41128.6 (−5878.9, −2378.2)
Proportion of out-of-pocket	Not select	40.8 (32.6, 48.9)	1377.3 (106.4, 168.3)

to improve health care value overall and address variation in health care costs in patients with CVD among low-cost and high-cost groups. It is particularly critical given the highly skewed distribution of health care costs with CVD in China. In previous studies, key factors of health care costs with CVD were often found using mean-based regression or for one of common diseases (9, 15, 19). To address the limitations, we used a machine learning with a principled backward stepwise algorithm (26) to identify determinants for varied levels of health care costs with CVD through a large survey with many covariates. We found that most of the key factors were the same across the quantiles, however, number of comorbidities was selected only the 10th percentile, while sex, age, medical payment and proportion of

self-payment were selected only the 90th percentiles. Through selecting the key factors based on various quantiles of health care costs and ranking the relative importance, our study showcased the a more appropriate method for a detailed understanding of how determinants explain the variability in different parts of the health care cost distribution.

One major contribution of our study was to quantify the drivers for health care costs in patient with CVD in different cost groups. Previous studies have confirmed the basic consensus that some factors that elevated the probability of being a high-cost users (28, 30, 32, 33). However, with only qualitative assumptions, the impact of potential drivers on changes in concentration remains ambiguous, and a rigorous quantitative assessment lacking. To bridge this gap, we examined the drivers of health care costs in patient with CVD using the quantile regression forests. Our results showed that if the proportion of inpatients with cardiomyopathy increased by 10%, the predicted Gini coefficient would increase by 2.11%, and the costs gap would increase 2.08%. This suggested that the low prevalence of serious diseases, represented by cardiomyopathy, but accompanied by high-cost characteristics, was an important driver of health care cost concentration. In recent years, the burden of disease in China has shifted considerably, with the epidemiological transition from acute diseases, to chronic disease, such as cardiovascular diseases (34). Furthermore, the prevalence of chronic diseases, and trends of specific chronic diseases, has increased (35). For example, cardiovascular hospitalization costs increased by more than 20% annually since 2004 (1), stroke prevalence increased by 155% and the incidence increased by 31.6% in rural areas from 1980s to 2013 (36). The prevalence and spending on cardiovascular diseases will continue to rise as China's demographics reflect population aging, prolonged life expectancy, increased expectation of medical care, and declining mortality rates, as well as the accumulation of risk factors.

We also found that these determinants did not uniformly impact the health care costs with linking the selected key determinants to health care costs using a weighted quantile regression. For example, number of comorbidities was selected only the 10th percentile. This finding might be surprising, given that it had been well documented that the high burden of comorbidities (37, 38). However, the magnitude of the relative difference was most profound at the lower percentile. It might be that the high cost of health care for those with CVD is more prominent among individuals with lower health care costs and has less of a differential impact on costs among those with higher costs and more complex conditions and care needs. In addition, medical payment method was selected only the 90th percentile. Compared with UEBMI, hospitalization costs were higher using the NRCMS/URBMI and lower with the full self-payment approach. This disproportionality in the effect estimates was often ignored in frequently used mean-based methods, potentially leading to biased conclusions.

Results from our study might also provide important insights for the development of tailored interventions to reduce potentially inappropriately high health care costs of CVD while maintaining or improving the care quality. For example, residence was significantly associated with higher percentile of health care costs of CVD. Due to the rich medial resources and top-notch medical technology, patients who seek medical treatment outside the city often suffer from more complex or severe diseases. Among the patients hospitalized in other provinces, 9.8% had Level-I medical procedure and 17.5% had Level-II medical procedure. These patients may be able to receive medical treatment locally. In addition, patients hospitalized in other provinces also incur more indirect costs, such as accommodation and transportation. Developing strategies by policymakers to reduce unnecessary or undesired treatments and related spending for out-of-town patients is warranted.

There are several limitations in the study. First, we conducted the large survey in almost all "AAA" general hospitals in S City, China. The findings generalized to the cities with rich medical resources but not the whole country. Second, we were not able to build causality in the relationships between health care costs and demographics, disease characteristics, and medical service utilization due to the cross-sectional nature of the survey data (39). However, our study identified determinants important for different quantiles of health care costs with CVD and can serve as a groundwork for future causal inference research in cost analysis. Third, we cannot evaluate other important variables that were not included in the study, either not measured or not collected in the survey, such as treatment quality or prognosis due to the lack of the follow-up. Despite the potential omitted variables, by using machine learning approach on a large sample that included individuals across demographics, residential information, comorbidity, surgery or operation information, insurance types, we believed our study deepens the understanding of the complex web of drivers and expands current research on CVD health care costs.

Conclusion

This study assessed the health care costs of treating all types of CVD in China and identified key determinants of high health care costs. To our knowledge, this is the first study assessing the economic burden of all types of CVD in China. Patients with high health care costs were more likely to be older, male, and have a longer length of hospital stay, more comorbidities, more complex medical procedures, and emergency admissions. Higher health care costs were also associated with specific CVD types such as cardiomyopathy, heart failure, and stroke. All of these findings may provide important insights for the development of tailored interventions to alleviate the financial burden of CVD in China, particularly among patients with high health care costs.

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 humans were approved by the institutional review board of School of Public Health at Shanghai Jiao Tong University School of Medicine on February 20, 2020 (IRB# SJUPN-202008). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

ML: Methodology, Validation, Writing – original draft. HG: Data curation, Resources, Writing – review & editing. CS: Writing – original draft. YX: Supervision, Writing – original draft. XL: Data curation, Writing – review & editing. LL: Writing – review & editing. YL: Conceptualization, Methodology, Writing – review & editing. GL: Funding acquisition, Project administration, Writing – review & editing.

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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.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1301276/full#supplementary-material

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Changes in characteristics of inpatient respiratory conditions from 2019 to 2021 (before and during the COVID-19 pandemic)

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Background: The COVID-19 pandemic has resulted in an increase in the number of individuals with respiratory conditions that require hospitalization, posing new challenges for the healthcare system. Recent respiratory condition studies have been focused on the COVID-19 period, with no comparison of respiratory conditions before and during the pandemic. This study aimed to examine hospital-setting respiratory conditions regarding potential changes in length of stay (LOS), mortality, and total charge, as well as socioeconomic disparities before and during the pandemic.

Methods: The study employed a pooled cross-sectional design based on the State Inpatient Data Nevada for 2019 (prior to the COVID-19 pandemic) and 2020–2021 (during the pandemic) and investigated all respiratory conditions, identified by the International Classification of Disease, 10th Revision codes ($n = 227,338$). Descriptive analyses were carried out for the three years. Generalized linear regression models were used for multivariable analyses. Outcome measures were hospital LOS, mortality, and total charges.

Results: A total of 227,338 hospitalizations with a respiratory condition were included. Hospitalizations with a respiratory condition increased from 65,896 in 2019 to 80,423 in 2020 and 81,018 in 2021. The average LOS also increased from 7.9 days in 2019 to 8.8 days in 2020 but decreased to 8.1 days in 2021; hospital mortality among patients with respiratory conditions increased from 7.7% in 2019 to 10.2% but decreased to 9.6% in 2021; and the total charges per discharge were $159,119, $162,151, and $161,733 from 2019 to 2021, respectively (after adjustment for the inflation rate). Hispanic, Asian, and other race patients with respiratory conditions were 1–3 times more likely than white patients to have higher mortality and LOS. Medicaid patients and non-White patients were predictors of a higher respiratory-related hospital total charge.

Conclusion: Demographic and socioeconomic factors were significantly associated with respiratory-related hospital utilization in terms of LOS, mortality, and total charge.

KEYWORDS

COVID-19, disparity, hospitalization, pandemic, respiratory

1. Introduction

The COVID-19 pandemic had a significant impact on hospitals all across the world. As the virus spread, hospitals quickly became overwhelmed, especially with patients requiring respiratory assistance and acute care (1–3). The pandemic has also highlighted existing healthcare disparities, with marginalized communities disproportionately affected by COVID-19 and faced barriers to accessing healthcare resources (4–6). The spectrum of respiratory conditions in the hospital setting has been previously compared before and during the pandemic (7). Comparing respiratory conditions in hospital settings before and during the pandemic in terms of mortality, length of stay (LOS), total charges, and sociodemographic characteristics requires more clarification and provides valuable insights into the effectiveness of healthcare systems in managing the pandemic and its associated respiratory burden.

Before the pandemic, studies have shown that older age, male gender, lower socioeconomic status, race, and certain comorbidities were associated with higher rates of hospitalization, LOS, and increased mortality among patients with respiratory conditions (8–10). However, the sociodemographic factors associated with hospitalization, LOS, and mortality changed during the COVID-19 pandemic (11, 12). COVID-19 disproportionately affected marginalized communities, including people of color (12–14), which might be due to a variety of factors, such as limited access to healthcare, social determinants of health, being essential workers (non-remote work), higher-density living environments, and underlying medical conditions (12, 15, 16). Comparing data from before and during the pandemic will reveal how to enhance patient care and formulate policies for future pandemics or other crises.

Despite the apparent disproportionate impact of the COVID-19 pandemic on communities, this impact varied across areas and over time (17–21). Evidence indicates that there is a significant difference in terms of disparity across the United States (17, 18). For example, COVID-19-related death disparity was highest among Hispanics in California (19), while it was highest among African Americans in New York (20), implying that social determinants of health can be different even in the same country, and local public health prevention strategies might be needed for equitable resource allocation. Evidence also indicates that the distinction between the early effects of the pandemic and its late effects can also be important from a policy standpoint, and it should be taken into account in investigations focusing on the COVID-19 pandemic (6, 17). It has been previously shown that emergency department visits for patients with mental illnesses increased throughout the COVID-19 pandemic in Nevada; however, this increase was significantly greater in the early pandemic in 2020 than in the late pandemic in 2021, implying the necessity of more resource allocations for mental health early in a pandemic (6). Data from the CDC also indicate that during 2020–2021, age-adjusted mortality decreased for African American (−6.1%), Asian (−1.9%), and Hispanic people (−1.2%) but increased for non-Hispanic Native Hawaiian or other Pacific Islander (68.3%), multiracial (57.1%), White (35.1%), and non-Hispanic American Indian or Alaska Native populations (3.8%) (21), providing robust evidence that the early impacts of the COVID-19 pandemic on the healthcare system could be different from its late impact.

In the present study, each year of the pandemic was individually investigated in descriptive analyses in order to distinguish the early effects of the pandemic from its late effects. The study period is from 2019 to 2021 (one year before the pandemic and two years during the pandemic). The aims of this study were to (1) compare the volume of inpatient care with respiratory conditions before and during the pandemic; (2) compare LOS, mortality, and total hospital charges of inpatient care with respiratory conditions before and during the pandemic; and (3) examine potential socioeconomic disparities in LOS, mortality, and total hospital charges of inpatient care with respiratory conditions in Nevada.

2. Methods

2.1. Data

The State Inpatient Data Nevada (SIDN), which contains all hospitalizations of community hospitals in Nevada, was used to collect inpatient data for 2019 (one year before the pandemic) as well as two years during the pandemic (2020 and 2021) to investigate an immediate impact of the pandemic in 2020 and a non-immediate impact in 2021. The database encompasses information on all non-federal acute community hospitals in Nevada. All hospitalizations associated with respiratory conditions were identified using the International Classification of Diseases, 10th Revision (ICD-10) (22). These codes are listed in Supplementary Table S1. The University of Nevada, Las Vegas, institutional review board deemed this study exempt (IRB# UNLV-2023-10) because the database provides administrative data after complete de-identification. For the data analysis, a total of 227,338 hospitalizations with respiratory conditions (2019–2021) were included in this study.

2.2. Measures and data analysis

The present study investigated three dependent variables: hospital LOS, mortality, and charges associated with a respiratory condition one year before the pandemic and two years during the pandemic. Age, gender, race/ethnicity, and payer source had previously been linked to these dependent variables (23) and were thus included as independent variables in the regression model. In order to control for time and detect a potential trend, year was included in all regression analyses, as used by other prior studies (6).

Multiple visits from the same patient would be considered distinct hospitalizations because the data had been deidentified. To account for variations within hospitals due to the clustering effect, we utilized the generalized linear model for multivariable analysis and treated hospitals as random effects while estimating the fixed effect of the independent variables of individual hospital discharges (6). The binomial family with the link function of logit was used for mortality analysis. The identity link function was used in linear regression for LOS and total charges. All statistical analyses were conducted using SAS software version 9.4 (SAS Institute Inc.; Cary, NC, USA). All p-values > 0.05 (2-tailed) were considered statistically significant.

3. Results

Table 1 indicates the characteristics of hospitalizations with respiratory conditions from 2019 to 2021. The frequencies of hospitalizations with a respiratory condition among all hospitalizations

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TABLE 1 Characteristics of hospitalizations with a respiratory condition in Nevada (2019–2021 SIDN).

Characteristic	2019	2020	2021	Value of *p*
N	65,896	80,423	81,018	
Age, mean (St.d.), years	61.2 (22.0)	61.9 (19.7)	61.3 (20.3)	<0.0001
Gender				<0.0001
Female	49.1%	46.3%	47.45	
Male	50.9%	53.7%	52.6%	
Race/ethnicity				<0.0001
White	66.3%	59.3%	59.1%	
Hispanic/Latino	8.1%	11.1%	10.3%	
African American	12.3%	12.2%	12.9%	
Asians	5.1%	6.3%	6.6%	
Others	6.2%	8.8%	8.7%	
Payer Status				<0.0001
Private insurance	22.0%	26.2%	27.5%	
Medicare	54.3%	49.5%	47.7%	
Medicaid	18.5%	17.9%	17.8%	
Self-pay	2.5%	2.8%	2.6%	
No charge	0.4%	0.3%	0.2%	
Others	2.5%	3.3%	4.1%	
Mortality	7.7%	10.2%	9.7%	<0.0001
Length of Stays (St.d.), days	7.9 (12.9)	8.8 (13.2)	8.1 (10.6)	<0.0001
Total charges per discharge	$159,119	$162,651	$161,733	<0.0001

SIDN, State Inpatient Data Nevada; St.d., standard deviation.

were 65,896, 80,423, and 81,018 from 2019 to 2021, respectively. In all of these three years, more than 50% of the hospitalizations were made by men, and their proportions were 50.9, 53.6, and 52.6% from 2019 to 2021, respectively (value of $p < 0.0001$). Medicare was the most prevalent payer source, but its percentage was reduced during the pandemic while the percentage for private insurance increased (value of $p < 0.0001$). The proportion of White people who were hospitalized significantly decreased from 66.3% in 2019 to 59.3% in 2020 and 59.1% in 2021, whereas it increased for other racial/ethnic groups, particularly for Hispanic and Asian people (value of $p < 0.0001$). The percentage of in-hospital mortality was 7.7% in 2019 and peaked at 10.2% in 2020. These differences reached statistical significance (value of $p < 0.0001$). LOS and total hospital charges per discharge were also significantly higher in 2020 and 2021 compared to 2019 (value of $p < 0.0001$). LOS from 7.9 days in 2019 peaked at 8.8 days in 2020. Total charges per discharge from $159,119 in 2019 peaked at $162,651 in 2020 (Table 1).

Table 2 indicates factors associated with in-hospital mortality among hospitalizations with respiratory conditions in Nevada from 2019 to 2021. Moreover, compared to 2019, 2020 (OR = 1.31; CI = 1.26–1.37; value of $p < 0.0001$) and 2021 (OR = 1.25; CI = 1.20–1.31; value of $p < 0.0001$) were associated with higher odds of in-hospital mortality. Age and gender were positively and negatively, respectively, associated with in-hospital mortality during these three years (Table 2). Compared

TABLE 2 Factors associated with mortality during hospitalizations with a respiratory condition in Nevada (2019–2021 SIDN[a]).

Independent variable	OR[b]	95% CI[c]	Value of *p*
Year*			
2019 (reference)			
2020	1.31	[1.26–1.37]	<0.0001
2021	1.25	[1.20–1.31]	<0.0001
Age*	1.40	[1.38–1.42]	<0.0001
Gender*			
Male (reference)			
Female	0.75	[0.72–0.77]	<0.0001
Race/Ethnicity*			
White (reference)			
African American	0.94	[0.90–0.99]	0.0399
Hispanic/Latino	1.13	[1.06–1.19]	<0.0001
Asian	1.21	[1.31–1.29]	<0.0001
Other races	1.28	[1.21–1.36]	<0.0001
Payer status*			
Private insurance (reference)			
Medicare	0.90	[0.86–0.94]	0.0112
Medicaid	1.03	[0.97–1.10]	0.3824
Self-pay	1.91	[1.74–2.09]	<0.0001
No charge	1.53	[1.14–2.07]	0.004
Others	1.27	[1.17–1.38]	<0.0001

[a]SIDN, State Inpatient Data Nevada. [b]Odds ratio, [c]Standard error. *$p < 0.0001$.

to the Whites, Hispanics (OR = 1.13; CI = 1.06–1.19; value of $p < 0.0001$) and Asians (OR = 1.21; CI = 1.31–1.29; value of $p < 0.0001$) with a respiratory condition had higher odds of in-hospital mortality during these three years. Compared to private insurance, self-pay (OR = 1.91; CI = 1.74–2.09; value of $p < 0.0001$) and no charge (OR = 1.53; CI = 1.14–2.07; value of $p < 0.0001$) were associated with higher odds of in-hospital mortality during these three years.

Table 3 indicates factors associated with LOS and total charges among hospitalizations with respiratory conditions in Nevada from 2019 to 2021 in a multivariable analysis. Compared to 2019, LOS in 2020 significantly increased. Hospital charges did not significantly increase either in 2020 or 2021 compared to 2019 (after controlling for the inflation rate). Age and gender were positively and negatively, respectively, associated with both LOS and total charges during these three years (Table 3). Compared to the White patients, Hispanic and Asian patients with a respiratory condition had both higher LOS and hospital charges during these three years. Medicare and self-pay patients were associated with lower LOS and total charges compared to those with private insurance, but Medicaid patients were associated with higher hospital charges and LOS during these three years.

4. Discussion

The present study investigated hospital utilization with a respiratory condition in Nevada from 2019 to 2021 (one year prior to

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TABLE 3 Factors associated with length of stay and total charges for hospitalizations with a respiratory condition in Nevada (2019–2021 SIDN[a]).

Independent variable	Parameter estimate	SE[b]	*p*-Value
Length of Stay			
Year*			
2019 (reference)			
2020	0.62893	0.064	<0.0001
2021	0.0641	−0.6	0.5495
Age*	0.65087	0.018	<0.0001
Gender*			
Male (reference)			
Female	−0.98544	0.051	<0.0001
Race/ethnicity*			
White (reference)			
Black	0.59159	0.081	<0.0001
Hispanic	1.01649	0.089	<0.0001
Asian	1.98345	0.109	<0.0001
Other races	3.32076	0.096	<0.0001
Payer status*			
Private insurance (reference)			
Medicare	−0.09295	0.068	0.1727
Medicaid	1.175	0.081	<0.0001
Self-pay	−1.81028	0.165	<0.0001
No charge	−0.88052	0.475	0.0637
Others	2.1665	0.149	<0.0001
Total charges			
Year*			
2019 (reference)			
2020	−2,095	1,336	0.1167
2021	−2,020	1,334	0.1299
Age*	12,497	367	<0.0001
Gender*			
Male (reference)			
Female	−27,726	1,067	<0.0001
Race/ethnicity*			
White (reference)			
Black	32,260	1,677	<0.0001
Hispanic	54,807	1,846	<0.0001
Asian	31,051	2,262	<0.0001
Other races	42,922	2,007	<0.0001
Payer status*			
Private insurance (reference)			
Medicare	−18,512	1,418	<0.0001
Medicaid	16,525	1,690	<0.0001
Self-pay	−48,662	3,429	<0.0001
No charge	−45,050	9,885	0.0012
Others	−27,026	3,092	<0.0001

[a]SIDN, State Inpatient Data Nevada. [b]SE, standard error. *$p < 0.0001$.

and two years during the COVID-19 pandemic) in terms of hospital LOS, mortality, and total charges. The proportions of hospitalizations with respiratory conditions were also examined. While previous studies focused on respiratory-related hospitalizations during the pandemic (24), the current study compared respiratory-related hospitalizations before the pandemic (2019) and during the pandemic period (2020 and 2021) to investigate the pandemic's disease and economic net burdens.

The results indicated a concerning trend in respiratory conditions in hospital settings during the COVID-19 period when compared to the pre-COVID-19 period. The frequency of hospitalization with respiratory conditions increased from 65,896 in 2019 to 81,018 in 2021 (Table 1), which may be attributed to the severe impact of COVID-19 pandemic on respiratory health or delayed healthcare seeking behavior due to the pandemic. The average LOS for patients with respiratory conditions also showed an increase. Prolonged hospital stays can put additional strain on healthcare resources (25). The reasons behind this longer LOS could be related to the complexity and severity of respiratory conditions during the pandemic, as well as potential challenges in providing timely and appropriate care (25). Moreover, it was found that the in-hospital mortality rate associated with a respiratory condition increased from 7.7% in the pre-COVID-19 period to 10.2 and 9.7% in 2020 and 2021, respectively (Table 1). The reasons for this increase could be multifactorial, including overwhelmed healthcare systems, resource constraints, and the increased severity of respiratory illnesses during the COVID-19 pandemic period (26). In addition to the increase in in-hospital mortality and LOS, the study highlighted a rise in total charges per discharge for patients with respiratory conditions. The increase in total charges during the COVID-19 pandemic period could be attributed to increased utilization of resources, higher demand for specialized care, longer LOS, and the additional expenses incurred due to infection prevention and control measures during the pandemic. It is worth mentioning that this increase did not reach statistical significance in the multivariable analysis (Table 3).

Current pandemic studies indicated that a higher proportion of hospitalizations due to COVID-19 were related to men, older adult, and racial/ethnic minority populations compared to their percentage in the population (17, 27, 28). Our study adds to the current literature indicating that during the pandemic period, compared to the pre-pandemic period, the proportion of males/females and non-Whites/Whites in hospitalized patients with a respiratory condition increased (Table 1). Moreover, our study identified certain demographic factors, including being older adult, male, and racial/ethnic minority groups, particularly Asians and Hispanics, that were associated with increased LOS, mortality and total charges for hospitalizations with respiratory conditions (Tables 2, 3). These findings raise concerns about racial/ethnic disparities and the need for targeted interventions to address the specific needs of vulnerable populations during both the COVID-19 and non-COVID-19 periods. Another explanation for our findings could be that male and racial/ethnic minority groups might underutilize hospice and palliative care services, a type of care that lower hospital LOS, charges, and mortality. Previous studies have indicated that male and racial/ethnic minority groups are less proactive about choosing a hospital palliative care referral or enrolling in hospice when their medical care is futile, potentially leading to longer hospital LOS and higher hospital charges and mortality rates (23, 29, 30). This mechanism is also applied to the Medicaid recipients (29).

The findings of our study are consistent with previous studies showing that race and ethnicity are related to hospital outcomes (17, 30), but Asian populations were the most vulnerable race/ethnicity in Nevada in terms of hospitalization with respiratory conditions. Asian populations are heterogeneous. Nevada has a sizable native Hawaiian community (31), an Asian population that has been frequently reported to struggle with health disparities (32). In contrast to Nevada, Hispanics and African Americans were the most vulnerable populations during the COVID-19 pandemic in California and New York, respectively (19, 20). The variation in the association of certain races/ethnicities with outcomes based on geographic region suggests that race/ethnic-centered localized policies are needed to address potential health disparities, particularly during respiratory-related pandemics.

This study had limitations that should be acknowledged. The study's reliance on retrospective data may introduce inherent biases and limitations in data quality, completeness, and accuracy. Additionally, the study focused solely on data from hospitals and did not account for healthcare services provided in other settings, such as outpatient care or home healthcare, which could lead to an underestimation of the true burden of respiratory conditions in the population. Moreover, the study highlighted disparities in outcomes based on race/ethnicity, gender, and age, but the underlying reasons for these disparities were not thoroughly investigated, warranting further research to better understand the contributing factors. Another limitation can be related to ICD codes, which are not error-free (23). These codes were used to identify respiratory conditions. However, due to our high sample size, these inaccuracies can only have a minor effect on how we interpret our results. Finally, data for 2022 were unavailable and were not included in the current analysis, which could have offered helpful information about respiratory conditions in the aftermath of the pandemic.

In conclusion, the findings of this study highlight the significant impact of the COVID-19 pandemic compared to pre-pandemic on hospital utilization for respiratory conditions. Demographic and socioeconomic factors were significantly associated with respiratory-related hospital utilization in terms of LOS, mortality, and total charge. These associations underscore the importance of implementing strategies to mitigate the burden on healthcare resources, improve patient outcomes, and address health disparities. Such strategies may include strengthening healthcare infrastructure, ensuring equitable access to care, and developing effective treatment protocols for respiratory conditions, particularly during times of crisis.

Data availability statement

The datasets presented in this article are not readily available because of the data user agreement between the authors and the data provider. Requests to access the datasets should be directed to the corresponding authors.

Ethics statement

The studies involving humans were approved by University of Nevada Las Vegas. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

ZM: Writing – original draft, Writing – review & editing. JY: Writing – review & editing. PK: Writing – review & editing. YK: Writing – review & editing. JS: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Software, Supervision, Validation, Writing – review & editing. B-LW: Conceptualization, Data curation, Formal analysis, Investigation, Supervision, Writing – review & editing.

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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.

Publisher's note

Supplementary material

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The cost-effectiveness of improved brief interventions for tobacco cessation in Thailand

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Background: This study estimated the cost-effectiveness of four strategies enhancing the quality and accessibility of Brief Intervention (BI) service for smoking cessation in Thailand during 2022–2030: (1) current-BI (status quo), (2) the effective-training standard-BI, (3) the current-BI plus the village health volunteers (VHV) mobilization, and (4) the effective-training BI plus VHV mobilization.

Methods: By interviewing five public health officers, nine healthcare professionals aiding these services, and fifteen BI service experts, we explored the status quo situation of the Thai smoking cessation service system, including main activities, their quantity assumptions, and activities' unit prices needed to operate the current cessation service system. Then, we modeled additional activities needed to implement the other three simulated scenarios. We estimated the costs and impacts of implementing these strategies over a nine-year operating horizon (2022–2030), covering 3years of service system preparation and 6years of full implementation. The modeled costs of these four strategies included intervention and program costs. The study focused on current smokers age 15years or older. The assessed impact parameters encompassed smoking prevalence, deaths averted, and healthy life-years gained. An Incremental Cost-Effectiveness Analysis compared the four simulated strategies was employed. Data analysis was performed using the One Health Tool software, which the World Health Organization developed.

Results: The findings of this investigation reveal that all three intervention strategies exhibited cost-effectiveness compared to the prevailing status quo. Among these strategies, Strategy 2, enhancing BI service quality, emerged as the most efficient and efficacious option. Therefore, the expansion of quality services should be synergistically aligned with augmented training, service delivery optimization, and managerial enhancements.

Conclusion: This approach is particularly poised to enhance accessibility to and the efficacy of smoking cessation interventions across Thailand.

KEYWORDS

tobacco cessation, economic evaluation, cost-effectiveness analysis, brief intervention (BI), Thailand

1 Introduction

Causes of death globally have evolved epidemiologically from infectious diseases to chronic degenerative diseases. In 2020, noncommunicable diseases (NCD) were responsible for 41 million deaths, which include eight million deaths from smoking (1, 2). For this reason, WHO has developed a global action plan for NCD prevention and control. One of the global targets is to reduce smoking rates by 30% between 2010 and 2025. The reduction of tobacco and other risk factors is necessary to achieve the Sustainable Development Goal (SDG) of reducing the number of premature deaths from NCD by 2030 (3, 4). In Thailand, smoking prevalence began to decline in 1991 from a peak of 32.0% down to 20.7% in 2009. Prevalence of smokers remained constant at around 19–20% until 2017 (5). The 2021 National Statistical Office's (NSO) Report on Tobacco Consumption Behavior survey showed that the prevalence of smoking in the Thai population age 15 years or older was 17.4%, with a prevalence of 34.7% among males and 1.3% among females (6). According to the 2015 National Health Survey in Thailand, two-thirds of smokers wanted to quit smoking, and 55% had tried, but only 7% were successful (7).

To effectively control tobacco consumption, WHO has developed the Framework Convention on Tobacco Control (FCTC), which has 168 signatory countries (8). The FCTC is operationalized through the MPOWER policy package. The MPOWER acronym stands for: (i) Monitoring tobacco uses and prevention measures, (ii) Protecting people from tobacco use, (iii) Offering cessation assistance, (iv) Warning about the dangers of tobacco, (v) Enforcing bans on tobacco advertising, and (vi) Raising taxes on tobacco (9). MPOWER has been shown to be effective in reducing tobacco use, shown by a reduction in the lung cancer mortality rate from 7 to 5% in Australia (10).

The "O" component of MPOWER refers to offering individual behavioral counseling, i.e., a brief intervention (BI) to help smokers quit smoking. For smoking cessation, the BI method consists of up to four face-to-face counseling sessions, with 5–10 min for each counseling session (11–13). The BI method increased the smoking cessation rate by an average of two percentage points of all current smokers within 1 year in the U.S. (14). In addition, a systematic review of 13 primary studies found that the BI achieved a success rate of 40% for quit attempts for its recipients (15, 16). This study estimated the cost-effectiveness of various hypothetical strategies aimed at enhancing the quality and accessibility of Brief Intervention (BI) service for smoking cessation in Thailand.

As mentioned earlier, BI has been shown to help smokers quit and, thus, the Thai health system has offered the BI service for smokers free of charge (17). In 2021, primary care providers verbally screened 15,281,599 people (28.8% of Thais age 15 years or over), of whom 9,921,801 said they were smokers. Providers offered smokers who wanted to quit help with quitting. However, only 1,548,287 smokers (15.6%) received help to quit tobacco use, and only 57,172 smokers (3.7%) who received BI successfully quit smoking (18). Those findings reflect two shortcomings of the Thai BI service system for smoking cessation: low access and poor efficacy.

In pursuit of enhancing the efficacy of Thailand's BI service system, a preliminary investigation was undertaken. This endeavor involved conducting interviews with pertinent stakeholders intimately involved in service provision. The selection criteria for these stakeholders emphasized substantial experience and affiliation with support systems, rendering their insights particularly valuable. The objective of the preliminary study was to comprehensively delve into the system's challenges and intricacies. Key stakeholders, including tobacco control experts, health providers, and officials from various administrative levels, were consulted to identify challenges in the current BI service system. The preliminary study identified two problems underlying low effectiveness: Inadequate training for BI providers resulting in a poor quality of BI services, and no effective strategy for improving access to these services. The researchers quantified the human, financial, and material resources, and macro-management support, that would be required at the national, regional, and provincial levels to implement a hypothetical BI strategy to increase smoker quit rates.

2 Materials and methods

In this study, we assessed the cost-effectiveness of candidate BI strategies for the current BI system in Thailand. The research focused on the "O" component of MPOWER, which reflects the policy of the Thai Ministry of Public Health (MOPH). The main health outcomes used to estimate BI effectiveness were smoking prevalence, the number of NCD-related deaths averted, and the number of Healthy Life-years (HLY) gained. The main program and intervention costs required for the implementation of each BI strategy were calculated. Program costs consist of human resources, as well as training, supervision, monitoring, evaluation, advocacy, infrastructure, general equipment, and overhead management costs. Intervention costs consist of the cost for healthcare to help smokers quit and BI specific supplies. Cost-effectiveness analysis of the different strategies was conducted using the incremental cost-effectiveness ratio (ICER) indicator (19).

2.1 Data collection and management

Acknowledging the paucity of conclusive evidence regarding the effect size of BI, our endeavor to enhance tobacco cessation BI strategies and ascertain their cost-effectiveness embraced a meticulously crafted data collection and management protocol, spanning four distinct phases: Exploration, strategy development, verification, and data analysis. This comprehensive approach involved soliciting insights from experts through a series of 15 key informant interviews. A diverse panel of experts with substantial experience in BI service systems was chosen to offer insights. These experts, drawn from varied service provision backgrounds, contributed valuable perspectives based on their deep understanding of analogous contexts. By amalgamating insights from multiple experts and conducting sensitivity analyses, the credibility of projections was significantly enhanced. While empirical data will eventually be collected, expert opinion plays a pivotal role in guiding the initial exploration and identifying potential effects. This iterative approach, combining expert insights with empirical evidence, results in a more nuanced and sophisticated analysis. The data collection phases are as follows.

In the exploration phase the current BI service system (i.e., the "status quo") was investigated. Target data for exploration included the BI service system's main activities, assumptions on quantity of interventions required, and calculation of the unit price for each

essential activity. An example of this would be for a BI provider who conducts four sessions of ten-minute interventions for each smoker, with a unit price of US$3 per smoker. Conducting the ten-minute BI service is the main activity, and a series of four BI sessions is the quantity assumption. In addition, a cost of $3 per smoker receiving this BI service is a unit price, which is calculated from the salary for 40-min equivalent of the nurse who providing the service.

In the strategy development phase, the researchers constructed hypothetical strategies for improving the BI service delivery system from the data obtained in the exploration phase. This study did not aim to design an alternative BI service *per se* but, rather, explored system improvements in order to more systematically deliver the internationally-recommended BI service.

In the verification phase, the main activities, quantity assumptions, and unit prices of each BI-improvement strategy were validated by the key informants.

During the final phase, encompassing data analysis, the researchers employed the OneHealth Tool (OHT) software. The OHT is specifically designed to facilitate strategic health planning, and was developed by the UN Inter-Agency Working Group. OHT is an indispensable resource for quantifying costing and technical support as part of the evaluation of the financial implications and health outcomes of interventions of interest. The costing framework encompasses personnel, training, monitoring/evaluation, and infrastructural overheads (e.g., water and electricity costs), formulated to project both cost dynamics and health impacts, inclusive of population growth, mortality reduction, and the diminishment of disease incidence or prevalence as intervention coverage expands (20–24). In the case of the present study, the indicators encompass rates of successful smoking cessation, averting pre-mature death, and the accrual of healthy life years (HLY).

2.2 Strategies to improve BI for smoking cessation

Based on the data collected from the exploration phase, we hypothesize four strategies to improve BI for smoking cessation. Each scenario delivers the BI service based on its capacity for the number of Thai smokers in 2021. For all four scenarios, the population of interest is Thais age 15 years or older residing in Thailand at the time of the study. The population in need of BI service are current smokers who reported smoking either regularly or occasionally within the past 12 months (i.e., 17.4% of the population age 15 years or older) (17).

We selected four BI-service system improvements based on two system-intervention approaches: Increasing BI service quality, and enhancing BI service accessibility. See Table 1 for details of the strategy descriptions, and their coverage rates and effectiveness.

The four strategies are arrayed as follows: Strategy 1: The status quo (no change in the approach that is currently being used by MOPH providers); Strategy 2: Implementing an effective training module to improve providers' quality in BI service; Strategy 3: Mobilizing community-based village health volunteers (VHV) to increase the accessibility to BI service for smokers; and Strategy 4: A combination of Strategies 2 and 3.

Strategy 1 represents the status quo (hereafter denoted as 'C-BI'). The C-BI service is offered in primary care settings nationwide, but is not systematically managed because only 3.5% of health care providers in the country have been trained to deliver the BI. Based on our preliminary assessment, five health personnel per province had received BI training, resulting in 385 trained people from 77 provinces, accounting for 3.5% of 10,666 primary healthcare providers in Thailand. The MOPH claimed that all of its healthcare providers offered the C-BI service, but many facilities did not have trained providers (25). Furthermore, the prevailing training package consists of only a two-day training workshop without any follow-up supervision. Therefore, many healthcare providers took an inordinate amount of time to offer the service and, thus, C-BI was not optimal or cost-effective. Based on the preliminary assessment, the providers spent 30 min per session talking about multiple topics instead of the required 5 min per session talking specifically about smoking cessation motivation. Hence, we defined Strategy 1 as a status quo situation providing the current, ineffective BI service, with ineffective training and non-systematic management. As the 'status quo' situation, providers verbally screen their adolescent and adult catchment population for smoking on an annual basis. Persons who admit to being smokers are offered help to quit tobacco use. This strategy's BI service covers 15.6% of Thai smokers and its effectiveness for quitting smoking is 3.7%.

Strategy 2 improves the BI service system by providing effective training for the BI providers in all primary healthcare settings to deliver standard BI service. Its acronym is 'ET-BI' which stands for effective training BI. In Strategy 2, the MOPH would offer a new BI training package that includes a two-day training workshop followed by two supervision sessions. The two-day training workshop would address the standard BI practice for smoking cessation (i.e., providing four, 5-min BI sessions per smoker). Trained BI providers would apply their real-world experience with the BI practices to consult with the trainers during two supervision sessions after the training to correct any errors. The MOPH would also train the BI providers working in all primary healthcare settings nationwide. During our verification phase, field experts confirmed that this approach could double the effectiveness of Strategy 1, from 3.7 to 7.4%.

Strategy 3 improves the BI service system by increasing smokers' access to BI over the C-BI service by mobilizing cadres of VHV to visit smokers in the community and encourage them to receive BI at the nearby primary healthcare provider. Its acronym is 'C-BI + VHV' standing for the current BI system plus the VHV approach. The primary healthcare system in Thailand consists of 9,766 sub-district health centers and 882,226 VHV (26). This is equivalent to 90 VHV per sub-district health center, and one VHV is assigned to take care of 10–15 households. In Strategy 3, the MOPH would mobilize 882,226 VHV nationwide to encourage smokers to seek BI service. Typically, VHV meet with the director of the local sub-district health center for 3 h every month to update their knowledge regarding community healthcare. In our Strategy 3, the VHV would be trained by the director of the sub-district health center to build skills in motivating smokers to use the BI service. The director would use video clips of best practices to train their cadre of VHV in a one-hour session, three times per year. The trained VHV would be expected to visit 15 smokers at least three times per year (if needed) to urge them to seek the local BI service. Based on our verification, field experts agreed that this VHV mobilization approach could double the BI accessibility rate from 15.6 to 33.2%.

Strategy 4 is a combination of Strategies 2 and 3: Effectively training BI providers to deliver the standard BI service (Strategy 2),

TABLE 1 Four BI strategies and hypothesized coverage and effectiveness rates.

Strategy	Acronym	Strategy description	BI coverage	BI effectiveness
Strategy 1	Status quo: current-BI (C-BI)	In the current situation in Thailand, 15.6% of smokers had access to cessation services, and 3.7% were in the process of quitting tobacco. Tambon Health Promoting Hospitals (THPH), district and provincial hospitals comprise only 3.5% of all MOPH health service facilities in Thailand. The providers have uneven capacity to deliver the BI, and most need to acquire more confidence in providing the BI, ensuring 30 min of tobacco cessation BI session.	15.6%	3.7%
Strategy 2	Effective-training standard-BI (ET-BI)	- Standard BI service (5 min per BI session); plus - New training package and systematic management	15.6%	7.4%
Strategy 3	Current-BI plus village health volunteers (VHV) mobilization (C-BI + VHV)	- Current BI service, and ineffective training and non-systematic management as in Strategy 1 plus - VHV mobilization	33.2%	3.7%
Strategy 4	Effective-training BI plus the VHV mobilization (ET-BI + VHV)	- Standard BI service, new training package and systematic management as in Strategy 2 plus - VHV mobilization	33.2%	7.4%

plus mobilizing VHV to screen and encourage smokers to seek BI service (Strategy 3). We hypothesize that Strategy 4 will have a smoking cessation success rate of 7.4% (similar to Strategy 2) and an accessibility rate of 33.2% (similar to Strategy 3).

For the purpose of this research, all four strategies were defined as nine-year interventions, to occur between 2022 and 2030. The first 3 years were designated as the preparation phase, which involves BI provider capacity building. During the six-year implementation phase, all strategies are applied at maximum capacity. In Strategies 2, 3, and 4, the target coverage rates for the first 3 years are defined as a progressive coverage increase from 50 to 80 to 100%, respectively. For Years 4 to 10, coverage is held constant at 100%. The effectiveness of each strategy varies depending on the quality of the capacity building, with Strategies 2 and 4 contributing a 7.4% effectiveness that is twice as large as Strategies 1 and 3 (3.7%). Table 2 shows a summary of the hypothesized coverage rates and effectiveness defined for each of the four BI strategies during the nine-year prediction interval.

2.3 Outcome measures

Outcome indicators of effectiveness of the four BI strategies include the prevalence of smoking, the number of NCD-related deaths averted, and the number of HLY gained. To calculate these outcomes, we employed the OHT parameters and estimation capacity based on a comprehensive literature review and meta-analysis (27–29) carried out by the OHT team at WHO. Hence, we use the default values of the OHT software, and that can be considered a limitation of this study. However, we updated the values of the prevalence rate based on the latest round (2021) of the Thai national Health Behavior of the Population survey. The parameters include total population, current smoking prevalence by age and sex, global-average relative risks of smoking for various NCD by age and sex (including ischemic heart disease, stroke, diabetes mellitus, asthma, chronic obstructive pulmonary disease, and cervical cancer), and attributable fractions (e.g., ischemic heart disease, stroke, diabetes mellitus, and chronic obstructive pulmonary disease). The OHT analysis providers an estimation of the strategy effectiveness by key parameters, relative risks, and attributable fraction in the population.

We entered the required data into the OHT software for analysis and projection of the effectiveness of the BI to quit tobacco use according to the outcome indicators. OHT provides the estimates of the gender- and age-specific prevalence of smoking (based on secular trends minus the impact of the intervention), the resulting numbers of NCD-related deaths averted by sex (assuming no other relevant interventions impacting mortality), and the number of HLY gained annually throughout the study period (2022-30).

2.4 Cost estimation of the interventions

The cost estimation for the BI system-improvement intervention was derived from the costs of the BI specific activities (i.e., the intervention cost in the OHT) and BI support activities (i.e., the program cost in the OHT). The program and intervention costs were the primary components of the total cost estimates. The program costs consist of the costs for human resources, training, supervision, monitoring, evaluation, advocacy, infrastructure, general equipment, and management. The intervention costs include the provider's time used to deliver the BI, and BI supplies (e.g., paper and pen).

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TABLE 2 Summary of BI coverage and effectiveness by strategy by year.

Strategy	Coverage	Intervention effectiveness (IE)		2022 (50%)	2023 (80%)	2024 (100%)	2025	2026	2027	2028	2029	2030
Strategy 1	15.6%	3.7%	Coverage	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%
			Impact (IE)	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%
Strategy 2	15.6%	7.4%	Coverage	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%	15.6%
			Impact (IE)	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%
Strategy 3	33.2%	3.7%	Coverage	24.4%	30.0%	33.2%	33.2%	33.2%	33.2%	33.2%	33.2%	33.2%
			Impact (IE)	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%	3.7%
Strategy 4	33.2%	7.4%	Coverage	24.4%	30.0%	33.2%	33.2%	33.2%	33.2%	33.2%	33.2%	33.2%
			Impact (IE)	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%	7.4%

We collected the cost data at macro and micro levels. Macro costing data were obtained from the Chairperson of the Division of Tobacco Product Control Committee, representatives from two MOPH regional offices for disease prevention and control, and representatives from two provincial health offices (PHO). The micro costing data were collected from healthcare service representatives from three provincial hospitals, representatives from three district hospitals, and representatives from three THPH.

The data in Supplementary Table S1 define the main BI activity.

Each year of Strategy 1 (status quo) includes ten core activities comprising the following: (1) Two meetings of the National Tobacco Control Board; (2) Eight meetings among staff of the Tobacco Control Division (under the Department of Disease Control, MOPH); (3) One policy instruction meeting between the Tobacco Control Division and PHO from all 77 provinces of Thailand; (4) A provincial policy cascade meeting among relevant health officers within each of the 77 provinces; (5) A three-day training of trainers (only for the first year); (6) A two-day BI training; (7) Two routine supervision sessions; (8) Print media (e.g., guidebook, infographics, posters, brochures); (9) Two monitoring and evaluation meetings between the Tobacco Control Division and the provincial level; and (10) Two monitoring and evaluation meetings for relevant health personnel within each province. Strategy 1 contains four 30-min BI sessions.

Strategy 2 has the same activities as Strategy 1, except that its BI training is a two-day workshop, two BI practice supervision sessions, and in-service education through an online learning platform for BI providers. Furthermore, its BI service contains four, 5-min BI sessions.

Strategy 3 has the same activities as Strategy 1, except that it has additional activities for VHV capacity-building, which includes three, 1-h learning sessions using five video clips, and learning through an online platform for VHV.

Strategy 4 combines the activities found in Strategies 2 and 3. The cost of VHV mobilization consists of function-related costs, e.g., training and annual meeting.

The calculation of indirect costs in all strategies involved the accounting of meal and transportation allowances for smokers. However, in Strategies 3 and 4, the indirect costs associated with Village Health Volunteers (VHV), such as labor and opportunity costs, were not factored into the cost calculation. This exclusion was justified by the assumption that the BI was integrated into routine VHV activities.

The questionnaires were systematically developed in accordance with the health costing frameworks established by the WHO within the OHT software (20). Unit prices for essential activities within the BI service domain were acquired through an expressly designed questionnaire survey. This survey was administered to 43 individuals directly involved in BI service activities, representing diverse settings and hierarchical levels within the healthcare system. These key informants include a person from the national level, three people from regional level, three people from the provincial level, four people from tertiary care hospitals, six people from secondary care hospitals, and 26 people from the primary care level. There are too many details regarding the main unit price calculation to describe fully in this article. The quantity assumptions and the unit prices of each core activity were input to calculate the unit cost values and, thereby, the intervention and program cost estimates (OHT 2019, when approximately 34 baht equaled one US dollar).

2.5 Cost-effectiveness analysis

We employed Incremental Cost-effectiveness Ratio (ICER), a fundamental metric in health economics used to compare the relative value of different healthcare interventions, to compare the cost-effectiveness of the four BI strategies (30). The calculation involved finding the cost and effectiveness differences between the interventions, then dividing the cost difference by the effectiveness difference. This yields the ICER, representing the additional cost incurred for gaining one additional unit of effectiveness. Decision-makers use ICER to evaluate the cost-effectiveness of interventions (by comparing the ratio to a predetermined threshold) in order to inform healthcare resource allocation decisions. In this study, we calculated the ICER in terms of US dollars per smoking quitter, NCD-related deaths averted, and per HLY gained for Strategies 2, 3, and 4 by using Strategy 1 as reference (Supplementary Table S2). Moreover, in order to quantify uncertainty, optimize decisions under diverse scenarios, and augment model credibility, a sensitivity analysis involving nine distinct strategies was conducted. The results of this analysis indicated no statistically-significant differences in effectiveness among the strategies examined (Supplementary Table S3). As a result, our study primarily focused on presenting four main strategies in its findings.

2.6 Ethical approval

The data collection tool and procedures complied with local and national regulations. Participants were informed of the purpose of the study. The study protocol was approved by the ethical committee of Institute for Population and Social Research, Mahidol University number; IPSR-IRB-2021-075. Service clients and the public WERE NOT involved in the design, or conduct, or reporting, or dissemination plans of our research.

3 Results

Table 3 provides a summary of the impacts of the four, nine-year BI-improvement interventions with respect to four health outcomes: Smoking prevalence, cumulative number of smoking quitters, cumulative number of NCD-related deaths averted, and the cumulative number of HLY gained. After the theoretical nine-year period of implementation, Strategies 2, 3 and, 4 provided better results than Strategy 1, with Strategy 4 performing the best. In Strategy 4, the smoking prevalence in 2030 is 16.6%, with an −0.8% absolute reduction in prevalence compared to 2021, when it was 17.6%. Over the 9 years (from 2022 to 2023) of observation, the cumulative number of smoking quitters, NCD-related deaths averted, and HLY gained were 251,108, 2,722, and 25,591, respectively.

Table 4 summarizes the results of the nine-year cost estimations for all four BI improvement strategies. Costs are presented in US dollars, and the discount rate, inflation, and depreciation rates were set at 0.0% in all periods due to unpredictable economic fluctuation during the Thai COVID-19 epidemic. The total costs were $64,065,591, $20,447,330, $129,497,192, and $40,191,330 for Strategies 1, 2, 3, and 4, respectively. Strategies 1 and 2 could each provide the BI service to 14,582,994 smokers because they applied the same 'business-as-usual' service accessibility strategy. Compared to Strategy 1, Strategy 2 has better health provider training, resulting in better BI service performance, but not better accessibility. Strategies 3 and 4 each have 29,753,800 smokers receiving the BI service because both employ the same VHV accessibility-enhancing strategy. From the data in Table 4, it can be seen that the intervention cost per smoker receiving BI service is equal between Strategies 1 and 3, as well as between Strategies 2 and 4. The reason for this is that the former pair applies the same business-as-usual, high-cost, ineffective BI service (e.g., 15 min per BI session), while the latter pair employs better training, and better BI performance (e.g., international standard 5 min per BI session). The program costs per smoker receiving BI service are $0.10, 0.12, 0.06, and 0.07, for strategies 1-4, respectively. As a result, the strategies with the lowest and highest costs-per-smoker receiving BI service are strategies 4, 2, 3, and 1, with $1.35, 1.40, 4.35, and 4.39, respectively. Based on the cost-per smoking-quitter provided by each strategy (Table 4), Strategy 4 has the cheapest unit cost ($160 per smoking quitter) and cost per NCD-related death averted ($14,765 per death averted).

Table 5 presents a summary of the estimated ICER per smoking quitter, NCD-related deaths averted, and HLY gained, for the four BI enhancement strategies. The study assumed periods of projection during the COVID-19 epidemic, and the discount, inflation, and depreciation rates were set at 0.0%. Based on the ICER, Strategies 2 and 4 cost less per additional unit of health outcomes compared to

TABLE 3 Projection effectiveness of BI strategy for tobacco cessation in Thailand.

Parameters	System-improvement intervention strategies			
	Strategy 1	Strategy 2	Strategy 3	Strategy 4
1. Smoking prevalence rate in 2030 (Prevalence in 2021 = 17.4%)	16.9%	16.8%	16.8%	16.6%
Males (prevalence at 2021 = 34,7%)	33.7%	33.5%	33.5%	33.0%
Females (prevalence at 2021 = 1.35%)	1.4%	1.4%	1.4%	1.4%
2. Absolute smoking prevalence change between 2021 and 2030	−0.5%	−0.6%	−0.6%	−0.8%
Males	−1.0%	−1.2%	−1.2%	−1.7%
Females	0.0%	0.0%	0.0%	0.0%
3. Cumulative number of smoking quitters	59,078	117,127	124,520	251,108
Males	56,480	112,515	119,933	241,147
Females	2,598	4,612	4,587	9,962
4. Cumulative number of NCD-related deaths averted	676	1,351	1,354	2,722
Males	544	1,089	1,093	2,197
Females	132	262	261	525
5. Cumulative number of HLY gained	6,420	12,868	12,746	25,591
Males	5,180	10,387	10,288	20,668
Females	1,240	2,481	2,458	4,923

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TABLE 4 Summary of costs by BI-improvement strategy for smoking cessation in Thailand (OHT 2019, 34 baht = 1 US dollar).

Parameters	Strategy 1	Strategy 2	Strategy 3	Strategy 4
1.Total cost	64,065,591	20,447,330	129,497,192	40,191,330
1.1 Intervention costs	62,587,292	18,677,066	127,704,868	38,107,040
1.2 Program costs	1,478,299	1,770,264	1,792,324	2,084,290
2. Number of smokers receiving BI service	14,582,994	14,582,994	29,753,800	29,753,800
3. Total cost-per-smoker receiving BI service	4.39	1.40	4.35	1.35
3.1 Intervention cost-per-smoker receiving BI service	4.29	1.28	4.29	1.28
3.2 Program cost-per-smoker receiving BI service	0.10	0.12	0.06	0.07
4. Total cost-per-smoking quitter	1,084.42	174.57	1,039.97	160.06
4.1 Intervention cost-per-smoking quitter	1,059.40	159.46	1,025.58	151.76
4.2 Program cost-per-smoking quitter	25.02	15.11	14.39	8.30
5. Total cost per NCD-related death averted	94,772	15,135	95,640	14,765

Discount, inflation, and depreciation rates were identified constantly at 0.0% during all projections due to unpredictable economic fluctuation during the Thai COVID-19 epidemic (31).

TABLE 5 Summary of estimated ICER per smoking quitters, NCD-related deaths averted, and HLY gained by BI-improvement strategy (OHT 2019, 34 baht = 1 US dollar).

Outcome	Strategy 1 = status quo (Comparator)		Strategy 2			Strategy 3			Strategy 4		
	Total cost	Total effects	Total cost	Total effects	ICER	Total cost	Total effects	ICER	Total cost	Total effects	ICER
Smoking quitter	64,065,591	59,078	20,447,330	117,127	−751	129,497,192	124,520	1,000	40,191,330	251,108	−124
Death averted	64,065,591	676	20,447,330	1,351	−64,620	129,497,192	1,354	96,507	40,191,330	2,722	−11,669
HLY gained	64,065,591	6,420	20,447,330	12,868	−6,765	129,497,192	12,746	10,343	40,191,330	25,591	−1,245

Strategy 1. Among the strategies for improving the BI system, Strategy 2 is the most efficient compared to the status quo (Strategy 1). That approach would save $751 per additional smoking quitter, $64,620 per additional death averted, and $6,765 per HLY gained when compared to the status quo. In addition, Strategy 4 would save $124 per additional smoking quitter, $11,669 per additional death averted, and $1, 245 per HLY gained when compared to the status quo.

4 Discussion

In order to provide MOPH policymakers with well-informed insights conducive to enhancing the cost-effectiveness of the BI service system, the present study undertook an analytical endeavor aimed at evaluating the postulated cost-effectiveness of four distinct BI strategies, delineated along two critical dimensions: augmentation of BI service quality and enhancement of BI service accessibility. The assessments encompassed perspectives from experienced consultants, alongside an examination of the outcomes associated with BI, notably within the context of smoking cessation initiatives. The findings provide compelling evidence for the potential of initiating substantial reforms in Thailand's tobacco-related BI strategies. The calculated Incremental Cost-Effectiveness Ratio (ICER) reveals that Strategy 2, characterized by effective training in BI service, emerges as the most economically viable approach for enhancing the BI service system when compared to the status quo (Strategy 1). The estimated total cost-per-smoking-quitter was approximately $175. This level of cost-effectiveness is lower than international benchmarks. For instance, in the United Kingdom, the cost per year of life saved through brief advice was £248 ($410), as reported in a prior study (32). Furthermore, another investigation indicated a cost of €354 per year of life saved ($495), derived from practitioners' brief advice within smoking cessation services. These comparative metrics underscore the significance of Strategy 2 as a financially prudent and impactful means of advancing tobacco-related intervention efforts (33). Note that all strategies are examined relative to Strategy 1, where the accessibility

of the current strategy is already factored in. Strategy 2 is the most cost-effective strategy for two reasons. First, Strategy 2 has a lower BI service cost because it provides four, 5-min BI sessions for each smoker instead of four 30-min BI sessions (as provided in the status quo strategy). In this strategy, the BI smoking cessation program was hypothetically provided to 60 smokers age 15 years or older. One-third (20 smokers) were projected to have quit smoking after 6 months of the program. This approach has been shown to be effective in helping smokers stay tobacco-free at 1 month (42.7% quitting rate) and at 6 months (35.0% quitting rate) after receiving the full BI service (34). Secondly, Strategy 2 has better BI training with the provision of two BI practice supervision sessions after the two-day BI training workshop and online learning platform. We modeled Strategy 2 based on a study that involved 626 senior nursing students. That study found that 91.7% of the participants felt that the training program enhanced their ability to assist BI clients to quit smoking. Well-designed 5A (e.g., Ask, Advise, Assess, Assist, Arrange) training courses could increase knowledge and self-efficacy of tobacco cessation among trained nurses (35). The government costs for improving BI service according to strategy 2 are around $20,447,330 for the 9 years, which is less than the cost of Strategy 1. Moreover, the online educational program and learning through BI practice were able to increase trainees' confidence and smoking cessation counseling skills. Students self-assessed higher levels of smoking cessation skills on Advising, Assessing, Assisting, and Arranging compared to baseline (36). Hence, the government costs for the improved BI service as per Strategy 4 are projected to be around $40,191,330 for the 9 years, which is less than Strategy 1.

The reason that Strategy 4 is the most effective, but not the most efficient, strategy, is because Strategy 4 broadens the accessibility of smoking cessation together with improving training, thereby incurring additional costs. The increased accessibility of Strategy 4 is based on Strategy 3 (the VHV- mobilization approach). Thailand has a long history of VHV involvement in assisting health personnel in service provision going back to the 1960s (37). Successful examples of VHV involvement in healthcare service provision include improvement in the nutritional status of children under the age of 5 years, household access to clean water, immunization coverage, expanded availability of essential drugs, as well as suicide prevention (38). There is evidence that VHV can be trained to assist smoking cessation in Thailand as well. A Thai quasi-experimental study in 2019 assessed the level of smoking-cessation-assistance ability among 64 VHV before and after participating in a program to boost health literacy for smoking cessation. That study found that the competency score after participating in the training program was significantly higher than the baseline score ($t = 2.78$, value of $p < 0.5$) (39). Thus, Strategy 4 is the best choice for achieving the global target of reducing smoking prevalence in Thailand through VHV training and involvement in smoking cessation provision. That said, Strategy 4 is less efficient in achieving additional quitters, NCD-related deaths averted, and HLY gained.

In sum, based on our findings, if the MOPH wishes to increase the effectiveness of the national BI-service system in the most cost-effective manner, they should adopt Strategy 2, which is an enhanced BI service system using effective training for primary healthcare personnel. If the government has the budget available, it could adopt Strategy 4, which is the most effective strategy for improving the BI service performance through the use of the effective BI training program and by involving VHV to increase accessibility to BI service.

Before integrating the study's findings into practical applications, it is crucial to address inherent limitations. One significant concern is the scarcity of research evidence, particularly regarding effect size, which necessitates careful interpretation. These limitations encompass various aspects, including the methods employed for data collection, expert opinions sought, and the choice of discount rate. Notably, data collection was confined to the perspective of the MOPH, emphasizing its specific role in BI services. Additionally, the evaluation of intervention costs considered only direct expenses, thus omitting crucial indirect costs from the analysis. Insights into service delivery heavily relied on estimates provided by healthcare professionals, which were validated by domain experts. Although the uniformity of discount and inflation rates employed in the analysis offers methodological convenience, it may not comprehensively capture the complexities of real-world dynamics. Therefore, the integration of the study's findings into practical contexts should be accompanied by a nuanced consideration of these limitations, ensuring a mindful and contextually-appropriate interpretation.

5 Conclusion

This study provides evidence that improvements to the national BI service system would be most cost-effective if Strategy 2 is applied (among four hypothetical scenarios). The study has demonstrated that the prevalence of tobacco use in Thailand could be reduce further by enhancing training and expanding access to BI for more smokers. The suggested changes to the current system for delivering BI services, as suggested in this study, could contribute to more efficient and effective health outcomes related to smoking cessation. Service delivery, access, and administration should be widened, and more effective service training should be incorporated.

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

This study protocol was approved by the ethical committee of Institute for Population and Social Research, Mahidol University number; IPSR-IRB-2021-075. Patients and the public were not involved in the design, or conduct, or reporting, or dissemination plans of our research.

Author contributions

RP: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Validation, Writing – original draft, Writing – review & editing. BS: Conceptualization, Formal analysis, Methodology, Supervision, Writing – original draft, Writing – review & editing. YS: Conceptualization, Supervision, Validation, Writing – review &

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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.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1289561/full#supplementary-material

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Trends and prescribing patterns of oral anti-neoplastic drugs: a retrospective longitudinal study

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Background: Cancer as a global public health problem, imposes a heavy disease burden. With the rapid development of oral anti-neoplastic drugs, there has been a paradigm shift in the treatment of cancer from intravenous to oral administration.

Objective: This study was conducted to investigate the trends and prescribing patterns of oral anti-neoplastic drugs in an academic tertiary hospital in China.

Methods: A single-center and retrospective analysis was performed based on the prescriptions of outpatients treated with oral anti-neoplastic drugs from 2017 to 2022. Yearly prescriptions and expenditure were calculated according to their pharmacological classes, and trends were further analyzed. Defined daily doses (DDDs) and defined daily cost (DDC) of oral targeted anti-neoplastic drugs were also determined.

Results: Both the number of prescriptions and expenditure of oral anti-neoplastic drugs increased progressively. There was a significant upward trend in the number and proportion of prescriptions for the older adult group, male group, and patients with gynecologic/genitourinary and respiratory cancer. Hormonal therapy agents accounted for the highest proportion of prescriptions, and letrozole was initially the most frequently prescribed drug. The number of DDDs of total oral targeted anti-neoplastic drugs showed a continuously ascending trend, primarily driven by the usage of epidermal growth factor receptor (EGFR) tyrosine kinase inhibitors (TKIs) and BCR-ABL TKIs.

Conclusion: The prescriptions and expenditure of oral anti-neoplastic drugs, and the number of DDDs of oral targeted anti-neoplastic drugs all showed a progressively ascending trend. Further studies are needed to evaluate the long-term health and financial outcomes, and the factors influencing these prescribing patterns.

KEYWORDS

prescribing patterns, oral anti-neoplastic drugs, outpatients, DDDs, DDC, EGFR-TKI

1 Introduction

Cancer is a substantial and global public health problem and is the second leading cause of death in the United States, contributing to a heavy burden of disease (1). Traditionally, the pharmacological therapy of cancer has predominantly been administered by intravenous route. However, oral anti-neoplastic drugs have experienced the highest rate of increase among all

anticancer medications over the past 13 years (2). Due to the rapid growth in the use of oral anti-neoplastic drugs, there has been a paradigm shift in the treatment of cancer, moving from intravenous to oral administration.

Compared with parenteral therapy, oral anti-neoplastic drugs offer several advantages, such as improved flexibility, better quality of life, and the absence of complications induced by invasive administration (3). However, this breakthrough treatment shifts the responsibility of appropriate administration and monitoring from healthcare professionals to the patients (4). Although oral anti-neoplastic drugs are preferred by patients, they have created new challenges regarding the effectiveness and safety, including reduced healthcare visits, patient medication adherence, regular monitoring and management of adverse events, and the risk of drug–drug interactions with other medicines and foods (5–8). Furthermore, many oral anti-neoplastic drugs, especially the targeted therapy agents, are costly and impose an increased financial burden (9). The Chinese government has implemented policies to cope with increasing prices and improve accessibility (10, 11).

Given the shift of cancer treatment paradigm and the increasing clinical importance, the patterns and trends of oral anti-neoplastic drugs have been studied in some countries over the past decade, mainly in France and Manitoba (9, 12). However, limited studies regarding this issue have been conducted in China, specifically focusing on certain classes of anti-tumor drugs, such as EGFR-TKIs and human epidermal growth factor receptor 2 inhibitors (11, 13). Therefore, the present study aims to investigate the prescribing patterns and trends of oral anti-neoplastic drugs at our institution in China from 2017 to 2022, and analyze the DDDs and DDC values of oral targeted anti-neoplastic agents during this time period.

2 Materials and methods

2.1 Study design

This was a retrospective and observational study that reviewed the outpatient prescriptions of Jinshan Hospital, Fudan University in China from January 1, 2017 to December 31, 2022. It is an academic tertiary hospital with 1,000 regular hospital beds. Ethical approval was obtained from the Ethical Committee of Jinshan Hospital, Fudan University (JIEC 2023-S77). The study was designed as a retrospective research and the data was extracted from prescription records, so informed consent was waived. Due to the single-center study design, the study population may not be representative of the general population, leading to biased results.

2.2 Data source and study population

All prescriptions for oral anti-neoplastic drugs for outpatients diagnosed with cancer were identified and obtained from the hospital information system between 2017 and 2022. Patients treated with oral anti-neoplastic drugs for non-oncologic indications and prescriptions with incomplete information were excluded from the analysis. The following prescription information was collected: prescription code, prescription date, clinical department, sex and age of the patient, diagnosis, drug generic name, route of administration, usage, single dosage, unit price, total amount and expenditure of the prescribed drug.

In accordance with the National Comprehensive Cancer Network (NCCN) guidelines, the classes of oral anti-neoplastic drugs were further classified into three categories as follows: (1) chemotherapy agents (capecitabine, hydroxycarbamide, tegafur gimeracil and oteracil porassium, chlorambucil, methotrexate, doxifluridine), (2) targeted therapy agents (gefitinib, erlotinib, osimertinib, almonertinib, imatinib, dasatinib, flumatinib, apatinib, anlotinib, olaparib, pyrotinib), (3) endocrine therapy agents (abiraterone, bicalutamide, flutamide, letrozole, tamoxifen).

2.3 Assessment of drug use

The primary units of analysis were the prescriptions and expenditure of oral anti-neoplastic drugs. The total number of yearly prescriptions containing oral anti-neoplastic drugs were counted and the percentage of prescriptions was calculated. The total and yearly costs were calculated by aggregating the costs of all the prescriptions in Chinese Yuan (CNY). The overall trends were illustrated by the annual number of prescriptions and annual expenditure. For further analyses, the trends were stratified by age group, sex, cancer type, drug classification and specific drug. Age groups were classified as young (0–39 years), middle-aged (40–64 years), and older groups (≥65 years).

The defined daily dose (DDD) is a statistical unit used for calculating and comparing medicine consumption. As there is no standard DDD for anti-neoplastic drugs, the DDD was obtained based on the daily dose and indications from the instructions and authoritative specification databases as well as literatures (13, 14). The defined daily doses (DDDs) were calculated as the ratio of the total dose of a specific drug used in grams to the corresponding DDD value. Higher DDDs values indicate a higher frequency of prescribing the medicine (15). The defined daily cost (DDC), as a standardized measure of the cost of per DDD medicine, was measured at the prescription level and recorded in Yuan. DDC was calculated with the following formula: DDC = expenditure/(number of DDDs) (16).

2.4 Statistical analysis

Data were processed via Microsoft Access software. Descriptive statistics were applied to characterize baseline patient demographics, cancer type, and the consumption of oral anti-neoplastic drugs. Continuous variables were presented by mean values and standard deviation. The frequencies and percentages per category were used to describe the categorical variables. The trends in yearly number and expenditure of prescriptions were analyzed by the Mann–Kendall trend test, and the trends in proportions were assessed by the log-linear test. All statistical analyses were conducted in R (4.1.0) software. Statistical significance was set at $p < 0.05$. Sankey diagrams of EGFR-TKIs in 2022 were plotted with the R package alluvial. The other figures were made using Prism 5.0 (GraphPad Software).

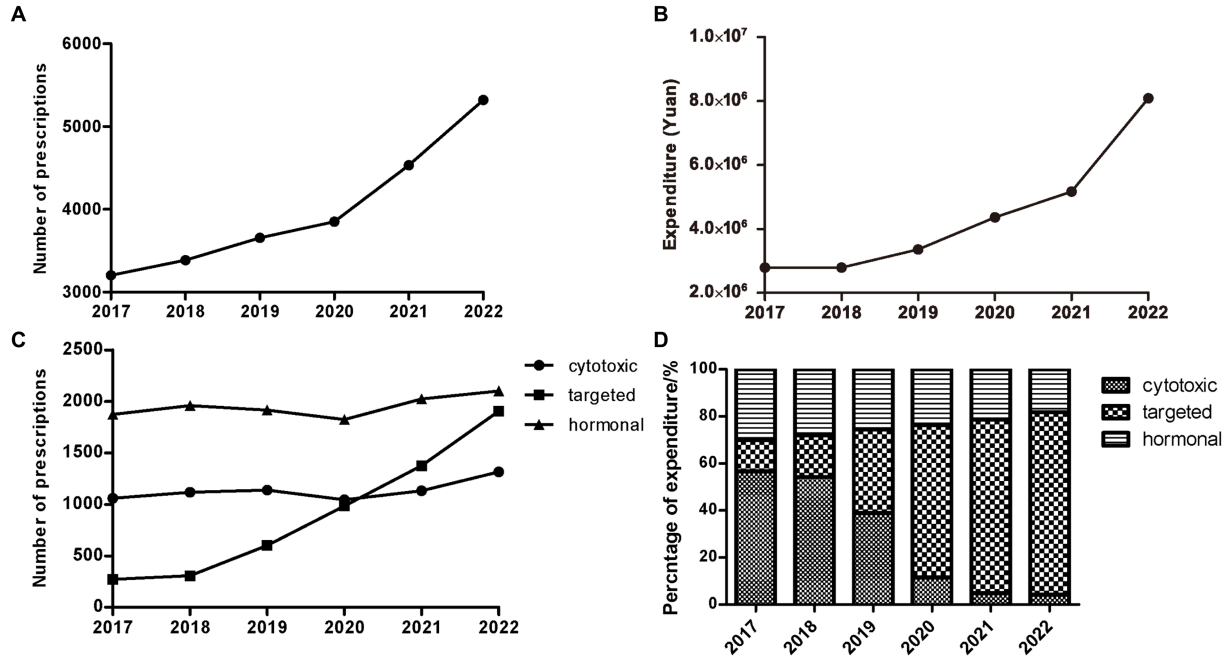

FIGURE 1
Trends of outpatients with treatment of oral anti-neoplastic agents from 2017 to 2022. **(A)** Trends of yearly total prescriptions. **(B)** Trends of yearly total expenditure. **(C)** Yearly prescriptions of different classes. **(D)** The percentage of yearly cost of specific drug classes.

3 Results

3.1 Overall trends in oral anti-neoplastic medication use and patient characteristics

Over the study period from 2017 to 2022, a total of 23,953 outpatient prescriptions meeting the inclusion criteria were included in the study. As shown in Figure 1A, the number of oral anti-neoplastic prescriptions increased progressively from 3,204 in 2017 to 5,322 in 2022, reflecting a 66.1% increase over the study period. The corresponding expenditure also continuously increased from 2,789,895 Chinese Yuan (CNY) in 2017 to 8,087,095 CNY in 2022 (Figure 1B). As Table 1 indicates, the average expenditure per prescription showed a significant increasing trend during the study period ($p = 0.024$).

The characteristics of patients with oral anti-neoplastic prescriptions are presented in Table 1. The average age of the population showed continuous growth from 63.5 years in 2017 to 67.1 years in 2021, but slightly decreased to 66.8 years in 2022. The number of prescriptions was concentrated in middle-aged patients aged 40–64 years and older adult patients aged ≥65 years. In the middle-aged group, the number of prescriptions remained stable ($p = 0.707$) and the proportion continuously decreased over the 6-year period ($p = 0.002$). However, both the number and percentage of prescriptions for the older adult group patients increased progressively (all $p < 0.01$). There was a significantly upward trend in the number and proportion of prescriptions for male (all $p < 0.01$). Conversely, the female patients showed a reduction in the proportion of prescriptions ($p = 0.001$), while exhibiting no clear trend in the number ($p = 0.26$).

With respect to cancer type, patients receiving oral anti-neoplastic drugs were frequently diagnosed with digestive tract, respiratory, breast, gynecologic/genitourinary and hematology cancer. Both the number and proportion of prescriptions increased significantly in gynecologic/genitourinary and respiratory cancer (all $p < 0.05$), whereas the proportion decreased in breast cancer over the study period ($p < 0.01$).

3.2 Trends in prescriptions stratified by drug class and individual drug

A total of 22 oral anti-neoplastic drugs in three drug categories were involved in the study. In terms of medication category, hormonal therapy agents accounted for the highest proportion of the total prescriptions, ranging from 39.48 to 58.49% (Figure 1C; Table 2). As shown in Figures 1C and D, the number of hormonal and cytotoxic agent prescriptions remained stable during the study period, and the percentage of expenditure exhibited downwards trends in both drug classes. However, the rapid growth was observed for targeted agent class, where the number of oral drugs available increased from three to eleven. The use of targeted agents increased rapidly from 271 prescriptions in 2017 to 1905 prescriptions in 2022, and the corresponding cost significantly increased from 13.12 to 77.51% (all $p < 0.05$).

The annual number of prescriptions and expenditure for oral anti-neoplastic agents by category and individual drug are presented in Tables 2, 3. Letrozole was the most frequently prescribed drug at the beginning of the study. However, proportion of prescriptions containing letrozole gradually declined from 34.43% in 2017 to 16.69% in 2022 ($p = 0.001$), and the corresponding expenditure decreased from 15.43 to 1.56% (all $p < 0.05$). In contrast, the percentage of prescriptions for EGFR-TKIs increased dramatically

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TABLE 1 Demographic and clinical characteristics of cancer patients treated with oral anti-neoplastic drugs between 2017 and 2022.

Variables	Number of patients (%)							P_1	P_2
	2017	2018	2019	2020	2021	2022	Total		
Age, years, mean ± SD	63.51 ± 12.75	63.66 ± 12.96	65.96 ± 12.85	66.40 ± 12.97	67.10 ± 12.49	66.81 ± 12.89			
Age group, n (%)									
Young (0–39)	95 (2.97)	124 (3.66)	110 (3.01)	109 (2.83)	107 (2.36)	158 (2.97)	703 (2.94)	0.707	0.237
Middle-aged (40–64)	1,600 (49.94)	1,633 (48.23)	1,442 (39.45)	1,480 (38.42)	1,596 (35.20)	1,826 (34.31)	9,577 (39.98)	0.707	0.002
Older adult (≥65)	1,509 (47.10)	1,629 (48.11)	2,103 (57.54)	2,263 (58.75)	2,831 (62.44)	3,338 (62.72)	13,673 (57.08)	0.009	0.005
Sex									
Male	1,189 (37.11)	1,286 (37.98)	1,609 (44.02)	1,853 (48.10)	2,265 (49.96)	2,703 (50.79)	10,905 (45.53)	0.009	0.002
Female	2015 (62.89)	2,100 (62.02)	2,046 (55.98)	1,999 (51.90)	2,269 (50.04)	2,619 (49.21)	13,048 (54.47)	0.260	0.001
Cancer type, n (%)									
digestive tract	638 (19.91)	731 (21.59)	828 (22.65)	668 (17.34)	782 (17.25)	970 (18.23)	4,617 (19.28)	0.133	0.170
Respiratory	55 (1.72)	47 (1.39)	269 (7.36)	520 (13.50)	759 (16.74)	1,152 (21.65)	2,802 (11.70)	0.024	0.006
Breast	1,450 (45.26)	1,465 (43.27)	1,341 (36.69)	1,178 (30.58)	1,260 (27.79)	1,223 (22.98)	7,917 (33.05)	0.133	<0.001
Gynecologic/ genitourinary	471 (14.70)	556 (16.42)	662 (18.11)	762 (19.78)	915 (20.18)	1,103 (20.73)	4,469 (18.66)	0.009	0.002
Hematology	569 (17.76)	577 (17.04)	548 (14.99)	683 (17.73)	743 (16.39)	811 (15.24)	3,931 (16.41)	0.060	0.292
Others	21 (0.66)	10 (0.30)	7 (0.19)	41 (1.06)	75 (1.65)	63 (1.18)	217 (0.90)	0.260	0.189
Total	3,204	3,386	3,655	3,852	4,534	5,322	23,953	0.009	
Average cost (CNY)	870.75	823.68	918.36	1132.13	1139.19	1519.56		0.024	

P_1, p-value for trend in number of prescriptions, assessed by Mann–Kendall trend test; P_2, p-value for trend in proportion of prescriptions, evaluated by log-linear analysis.

from 0.24% in 2018 to 17.44% in 2022 ($p = 0.014$), and the corresponding cost also substantially rose from 2.45 to 43.73% ($p = 0.012$). Consequently, by the end of the study, EGFR-TKIs had become the most commonly prescribed drugs, with the highest expenditure.

3.3 DDDs and DDC values of oral targeted anti-neoplastic agents

The number of DDDs of all oral targeted anti-neoplastic drugs exhibited a continuously ascending trend over the study period (Figure 2A). The average DDC of all targeted drugs changed slightly from 2017 to 2019 and from 2020 to 2022, but increased significantly from 2019 to 2020 due to the availability of olaparib and pyrotinib (Figure 2B).

As shown in Figure 2C, EGFR-TKIs and BCR-ABL TKIs were the most widely applied drugs, accounting for the majority of the total DDDs of targeted drugs from 2019 to 2022. The DDDs value of EGFR-TKIs increased dramatically from 2018 to 2022, and the DDDs value of BCR-ABL TKIs increased gradually since 2017. At the end of study, the number of DDDs of EGFR-TKIs was far more than that of other targeted drugs. However, the average DDC values of EGFR-TKIs and BCR-ABL TKIs were both at relatively low levels, and decreased year by year from 2017 to 2021 and 2017 to 2022, respectively. In addition, the DDC of the other targeted drugs all showed a descending trend (Figure 2D).

3.4 The consumption and distribution of EGFR-TKIs

The distribution of DDDs and DDC values of drugs belonging to EGFR-TKIs in each year is shown in Figure 3, including gefitinib, erlotinib, osimertinib, and almonertinib. The number of DDDs of all four drugs increased dramatically, while the DDC of gefitinib and osimertinib decreased substantially in 2019 and 2021, respectively. The DDC of erlotinib and almonertinib remained relatively unchanged in 2021 and 2022. The Sankey diagram illustrated the relationship between age group, sex and the distribution of EGFR-TKIs in 2022 through lines, signifying the quantities via the line width (Figure 4).

4 Discussion

Oral anti-neoplastic therapy has emerged as a novel paradigm of anti-tumor treatment in recent years. The present study described the real-world patterns and trends regarding the use of oral anti-neoplastic drugs in a tertiary hospital in China over a 6-year period. Both the yearly prescriptions and corresponding expenditures of oral anti-neoplastic drugs increased progressively throughout the study period, particularly for oral targeted anticancer drugs. The DDDs of overall oral targeted anti-neoplastic drugs showed a continuously ascending trend over the study period, with EGFR-TKIs and BCR-ABL TKIs accounting for the largest proportion. In line with previous research conducted in other countries, this study found a significant increase

TABLE 2 Number of oral anti-neoplastic prescriptions from 2017 to 2022.

Medicine	Number of prescriptions (%)							P_1	P_2
	2017	2018	2019	2020	2021	2022	Total		
Cytotoxic class	1,059 (33.05)	1,118 (33.02)	1,139 (31.16)	1,044 (27.10)	1,132 (24.97)	1,316 (24.73)	6,808 (28.42)	0.260	0.002
Capecitabine	37 (1.15)	110 (3.25)	254 (6.95)	319 (8.28)	432 (9.53)	505 (9.49)	1,657 (6.92)	0.009	0.018
Hydroxycarbamide	278 (8.68)	289 (8.54)	301 (8.24)	349 (9.06)	383 (8.45)	379 (7.12)	1,979 (8.26)	0.024	0.210
Tegafur gimeracil and oteracil porassium	524 (16.35)	580 (17.13)	481 (13.16)	310 (8.05)	272 (6.00)	373 (7.01)	2,540 (10.60)	0.133	0.008
Chlorambucil, methotrexate, and doxifluridine	220 (6.87)	139 (4.11)	103 (2.82)	66 (1.71)	45 (0.99)	59 (1.11)	632 (2.64)	0.024	0.001
Targeted class	271 (8.46)	308 (9.10)	600 (16.42)	984 (25.55)	1,377 (30.37)	1,905 (35.79)	5,445 (22.73)	0.009	0.001
EGFR-TKIs	13 (0.41)	8 (0.24)	194 (5.31)	347 (9.01)	523 (11.54)	928 (17.44)	2,013 (8.40)	0.024	0.014
BCR-ABL TKIs	258 (8.05)	295 (8.71)	319 (8.73)	401 (10.41)	412 (9.09)	511 (9.60)	2,196 (9.17)	0.009	0.113
Apatinib, anlotinib, olaparib, and pyrotinib	0 (0.00)	5 (0.15)	87 (2.38)	236 (6.13)	442 (9.75)	466 (8.75)	1,236 (5.16)	0.008	0.056
Hormonal class	1,874 (58.49)	1,960 (57.89)	1,916 (52.42)	1,824 (47.35)	2,025 (44.66)	2,101 (39.48)	11,700 (48.85)	0.260	<0.001
Letrozole	1,103 (34.43)	1,140 (33.67)	1,129 (30.89)	918 (23.83)	967 (21.33)	888 (16.69)	6,145 (25.66)	0.133	0.001
Tamoxifen	339 (10.58)	307 (9.07)	177 (4.84)	191 (4.96)	212 (4.68)	200 (3.76)	1,426 (5.95)	0.452	0.008
Bicalutamide	334 (10.42)	381 (11.25)	477 (13.05)	584 (15.16)	608 (13.41)	626 (11.76)	3,010 (12.57)	0.009	0.306
Abiraterone and flutamide	98 (3.06)	132 (3.90)	133 (3.64)	131 (3.40)	238 (5.25)	387 (7.27)	1,119 (4.67)	0.060	0.031

EGFR-TKIs, gefitinib, erlotinib, osimertinib, and almonertinib.
BCR-ABL TKIs, imatinib, dasatinib, and flumatinib.
P_1, *p*-value for trend in number of prescriptions, assessed by Mann–Kendall trend test; P_2, *p*-value for trend in proportion of prescriptions, evaluated by log-linear analysis.

of 66.1% in overall prescriptions and a 1.90-fold increase in expenditure (12, 17). A retrospective population-based study conducted in Manitoba demonstrated that the prevalence of oral anticancer agents use increased from 222 per 100,000 to 328 per 100,000, and the total cost of targeted oral anticancer agents per year for all cancer patients increased from $1.8 million to $19 million between the years 2003 and 2016 (12). Similarly, a retrospective analysis using survey data in France revealed that the proportion of cancer patients receiving oral anticancer treatments increased by four percentage points from 2004 to 2012, with an increase from 28.4 to 32.5% (17). This trend may reflect changes in cancer incidence and diagnosis patterns over time (12). Furthermore, the availability of oral anti-neoplastic drugs has significantly increased in recent years, leading to a shift toward the adoption of them as a therapeutic strategy for certain cancers (18). In terms of drug categories, the prescriptions of oral targeted anti-neoplastic drugs showed a significant increase during the study period. The rapid increase in prescriptions for targeted agents can be attributed to several factors. Firstly, with the advancements in medical technology and research, FDA-approved targeted agents have experienced a remarkable surge in the past 20 years and have become mainstream cancer treatments (19). Secondly, targeted drugs have shown high potency, reduced toxicity and increased survival rates compared to traditional chemotherapy drugs. This has resulted in a growing interest and demand for these treatment options among healthcare providers and patients. Thirdly, the accessibility and affordability of targeted anti-cancer drugs have significantly increased following the implementation of government healthcare policies (20, 21). The prescriptions of oral cytotoxic and hormonal anti-neoplastic drugs remained stable, resulting in decreasing proportions.

In this study, the average age of the population increased gradually from 2017 to 2021, and both the number and percentage of prescriptions rose progressively in older adult patients aged ≥65 years. These results may be attributed to increased life expectancy, longer survival of patients and preference of clinicians for oral anticancer drugs instead of parenteral therapy in older adult patients to improve their quality of life (9). The absence of social support, limited access to care, and financial constraints may serve as major obstacles for older patients in utilization of oral anticancer drugs (22).

In the first few years of this study, the number of prescriptions for females was dramatically greater than those for males due to the high proportion of letrozole. Letrozole is known as a potent third-generation aromatase inhibitor and has been widely used in the adjuvant, neoadjuvant, and metastatic therapy of hormone receptor-positive breast cancer (23). However, with the widespread application of oral targeted anticancer drugs in clinical practice, the disparity in proportion between males and females has gradually decreased.

Regarding oral targeted anti-neoplastic drugs, the number of DDDs showed an upward trend, while the number of DDC showed a downward trend. DDC is an efficient indicator that guides the market prices of pharmaceutical products (15). Consistently, previous studies have indicated that drug price was an important determinant of drug consumption, and there was a negative relationship between DDC and the number of DDDs (11, 13, 24). In order to improve the availability and affordability of drugs, China established its fundamental health insurance system in 2009, which was enhanced by the implementation of price negotiation and mandatory reimbursement policies in 2017 (13). The policies of price negotiation have been implemented in many countries, which resulted in a significant reduction in drug prices and an increase in drug consumption (24, 25). The Chinese government

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TABLE 3 Cost of oral anti-neoplastic drugs from 2017 to 2022.

Medicine	Cost of Chinese yuan (%)							P_1	P_2
	2017	2018	2019	2020	2021	2022	Total		
Cytotoxic class	1,584,579 (56.80)	1,511,733 (54.20)	1,309,118 (39.00)	510,662 (11.71)	255,695 (4.95)	348,882 (4.31)	5,520,669 (20.79)	0.024	0.002
Capecitabine	27,724 (0.99)	74,766 (2.68)	159,188 (4.74)	179,518 (4.12)	115,736 (2.24)	120,759 (1.49)	677,691 (2.55)	0.260	0.818
Hydroxycarbamide	14,559 (0.52)	14,010 (0.50)	14,686 (0.44)	18,249 (0.42)	19,934 (0.39)	38,206 (0.47)	119,644 (0.45)	0.024	0.187
Tegafur gimeracil/ oteracil porassium	1,450,872 (52.00)	1,374,210 (49.27)	1,094,723 (32.61)	290,648 (6.66)	113,203 (2.19)	179,125 (2.21)	4,502,781 (16.96)	0.024	0.003
Chlorambucil, methotrexate, and doxifluridine	91,424 (3.28)	48,747 (1.75)	40,521 (1.21)	22,247 (0.51)	6,822 (0.13)	10,792 (0.13)	220,553 (0.83)	0.024	<0.001
Targeted class	366,015 (13.12)	495,570 (17.77)	1,188,343 (35.40)	2,812,015 (64.48)	3,801,939 (73.61)	6,268,651 (77.51)	14,932,533 (56.25)	0.009	0.003
EGFR-TKIs	68,418 (2.45)	45,677 (1.64)	535,537 (15.95)	1,039,834 (23.84)	1,528,017 (29.58)	3,536,276 (43.73)	6,753,759 (25.44)	0.024	0.012
BCR-ABL TKIs	297,597 (10.67)	423,309 (15.18)	391,233 (11.66)	487,121 (11.17)	534,769 (10.35)	721,350 (8.92)	2,855,379 (10.76)	0.024	0.176
Apatinib, anlotinib, olaparib, and pyrotinib	0 (0.00)	26,584 (0.95)	261,573 (7.79)	1,088,808 (24.97)	1,724,705 (33.39)	1,873,119 (24.87)	5,323,395 (20.05)	0.008	0.064
Hormonal class	839,301 (30.08)	781,680 (28.03)	859,153 (25.60)	1,038,287 (23.81)	1,107,468 (21.44)	1,469,562 (18.17)	6,095,451 (22.96)	0.024	<0.001
Letrozole	430,374 (15.43)	414,166 (14.85)	389,613 (11.61)	334,802 (7.68)	102,512 (1.98)	125,783 (1.56)	1,797,250 (6.77)	0.024	0.006
Tamoxifen	9,115 (0.33)	7,120 (0.26)	36,991 (1.10)	48,020 (1.10)	54,488 (1.05)	64,876 (0.80)	220,610 (0.83)	0.024	0.115
Bicalutamide	364,249 (13.06)	318,797 (11.43)	389,251 (11.60)	563,784 (12.93)	442,840 (8.57)	360,276 (4.45)	2,439,197 (9.19)	0.707	0.059
Abiraterone and flutamide	35,563 (1.27)	41,597 (1.49)	43,298 (1.29)	91,681 (2.1)	507,628 (9.83)	918,627 (11.36)	1,638,394 (6.17)	0.008	0.017

EGFR-TKIs, gefitinib, erlotinib, osimertinib, and almonertinib.
BCR-ABL TKIs, imatinib, dasatinib, and flumatinib.
P_1, *p*-value for trend in number of prescriptions, assessed by Mann–Kendall trend test; P_2, *p*-value for trend in proportion of prescriptions, evaluated by log-linear analysis.

has implemented six rounds of national drug price negotiations since 2016 to include innovative and expensive drugs, particularly anticancer drugs, in the national list of reimbursable medicines. The average price discount for newly-added drugs has exceeded 50% from 2018 to 2021 (26). Furthermore, national centralized drug procurement organized by the Chinese government has effectively reduced the price of bid-winning anti-cancer drugs (27). National volume-based procurement was launched in 2018 to negotiate drug price with manufacturers in "4+7" pilot cities, which covered 4 provincial municipalities and 7 sub-provincial cities (10). The procurement volume was set based on 60–70% of the actual annual drug consumption of all public hospitals in pilot cities in the previous year (21). The centrally purchased pilot drugs were high-quality generic drugs with the same therapeutic effect through the quality consistency evaluation procedure. Most of them were drugs for the treatment of chronic diseases, and major diseases such as tumors.

In this study, the DDDs value of EGFR-TKIs increased dramatically from 2018 to 2022, far exceeding that of other targeted drugs at the end of the study. The latest monitoring data in 2023 showed that lung cancer had been the leading cancer in Shanghai. EGFR-TKIs have been used as a novel therapeutic strategy in patients with advanced non-small cell lung cancer (NSCLC) with EGFR mutations, significantly improving prognosis. Although the first generation EGFR-TKIs, including gefitinib and erlotinib, have shown high efficacy, resistance has emerged due to T790M mutation (28). Osimertinib, a third-generation and central nervous system-active EGFR-TKI targeting T790M mutation, has exhibited longer progression-free survival (PFS) compared to the first generation EGFR-TKIs (29). Almonertinib, another third-generation EGFR-TKI, was approved by China in March 2020 as a novel treatment option for EGFR T790M+ NSCLC (30). Our data showed a marked increase in the number of DDDs for gefitinib and osertinib, far exceeding erlotinib and almonertinib. Geftinib and erlotinib have been included in the national health insurance in 2017, and osimertinib in 2019. Additionally, multiple rounds of price negotiations have been conducted for EGFR-TKIs, resulting in a decrease in DDC and improved availability (11).

The results of the present study showed that both the number of BCR-ABL TKIs prescriptions and expenditure significantly increased over the study period. Moreover, the DDDs value of BCR-ABL TKIs

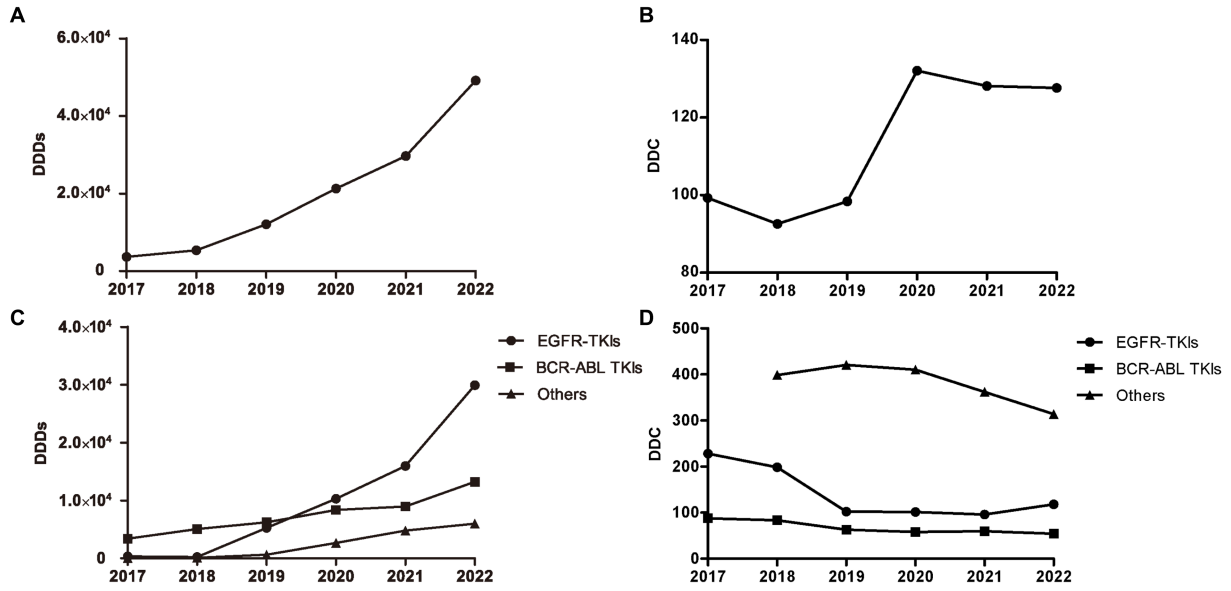

FIGURE 2
DDDs and DDC values of oral targeted anti-neoplastic drugs from 2017 to 2022. **(A)** DDDs of overall targeted anti-neoplastic drugs. **(B)** DDC of overall targeted anti-neoplastic drugs. **(C)** DDDs of three classes. **(D)** DDC of three classes. DDDs, defined daily doses; DDC, defined daily cost.

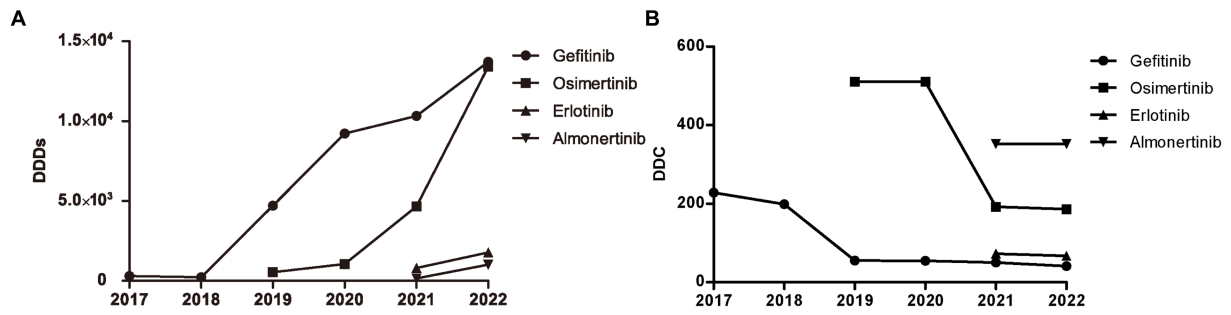

FIGURE 3
Consumption of four types of EGFR-TKIs from 2017 to 2022. **(A)** DDDs of four types of EGFR TKIs. **(B)** DDC of four types of EGFR TKIs. DDDs, defined daily doses; DDC, defined daily cost.

has been gradually increasing since 2017, accompanied by a gradual decline in DDC. BCR-ABL TKIs are the first-line treatment for patients with chronic myelocytic leukemia (CML) and Ph chromosome-positive acute lymphoblastic leukemia (31). With the development of BCR-ABL TKIs including first-generation imatinib and second-generation dasatinib, most CML patients experience long-term remissions and have a life expectancy close to normal (32). Flumatinib, a novel second-generation BCR-ABL TKI, has been launched in the Chinese market with an optimized structure based on imatinib to achieve higher potency (33).

There were certain limitations in our study. First, it was a single-center and retrospective design, which restricted the generalizability of the data to other populations or settings. Second, the term consumption referred to the quantity of prescribed drugs rather than administered drugs, due to the lack of information about medication compliance. Third, our analysis was carried out based on prescription data, and lacked the clinical information such as laboratory tests and biological characteristics, which limited the ability to assess the appropriateness of drug prescriptions and evaluate factors influencing prescribing patterns. Furthermore, while the study focused on prescription trends and drug consumption, it does not provide insights into patient outcomes or the effectiveness of the prescribed treatments.

5 Conclusion

In this study, the use of oral anti-neoplastic drugs has been increasing annually, especially oral targeted anticancer drugs. Among them, EGFR-TKIs and BCR-ABL TKIs accounted for the majority in terms of defined daily doses (DDDs), but with relatively low levels of DDC. Further studies are warranted to explore the factors influencing the prescribing patterns and to conduct the cost-effectiveness analysis of oral anti-neoplastic treatments.

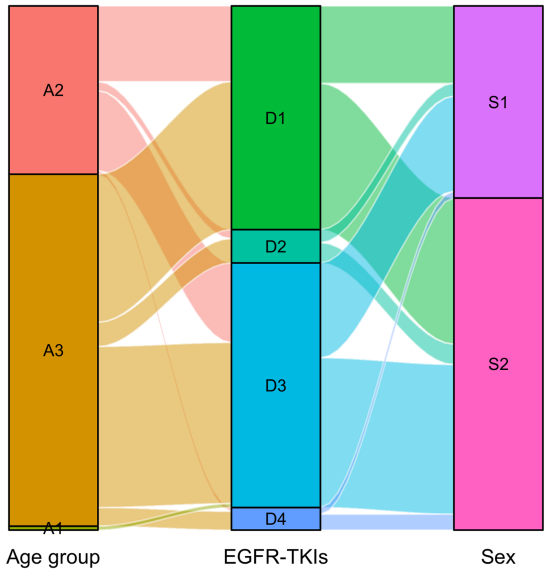

FIGURE 4
Sankey diagram of EGFR-TKIs in 2022. The age group and sex matched with the distribution of EGFR-TKIs. A1: Young, A2: Middle-aged, A3: Older group, D1: Gefitinib, D2: Erlotinib, D3: Osimertinib, D4: Almonertinib, S1:Male, S2: Female.

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 humans were approved by Ethics Committee of Jinshan Hospital, Fudan University. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

XL: Conceptualization, Funding acquisition, Investigation, Writing – original draft, Writing – review & editing. WR: Data curation, Resources, Validation, Writing – original draft. SR: Methodology, Project administration, Writing – original draft. YZ: Data curation, Project administration, Writing – original draft. JZ: Methodology, Software, Writing – original draft. JC: Formal analysis, Software, Visualization, Writing – review & editing. NZ: Conceptualization, Resources, Supervision, Validation, Writing – review & editing.

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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.

Publisher's note

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Spatiotemporal evolution of healthcare service capacity at township health centers in China

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Introduction: This study analyzes the efficiency, spatiotemporal evolution, and influencing factors of provincial township health centers' healthcare service capacity in China.

Method: It utilizes an unexpected output super-efficiency slacks-based measure (SBM) model, exploratory spatiotemporal data analysis methods, and a quantile regression model.

Results: The results show that the healthcare service capacity of township health centers is better in provinces with a larger proportion of hierarchical diagnoses and treatments pilot projects in cities, and the regional efficiency trend is ordered central > eastern > western > northeastern. The healthcare service capacity of provincial township health centers mainly shows significant spatial correlation and a spatiotemporal distribution pattern of "high agglomeration, low differentiation."

Discussion: Rural population density and per capita GDP significantly improve the healthcare service capacity of township health centers, while local governments' healthcare and health expenditure increases the healthcare service capacity of township health centers in certain quantiles. The urbanization rate and per capita disposable income inhibit the improvement of the healthcare service capacity of township health centers in certain quantiles. The provinces should accelerate the promotion of hierarchical diagnoses and treatment pilot projects in cities and establish national cooperative development models to promote public health.

KEYWORDS

Township health center, healthcare service capacity, unexpected output superefficiency SBM, exploratory spatiotemporal data analysis, quantile regression

1 Introduction

The 19th National Congress of the Communist Party of China proposed the construction of a "Healthy China" by promoting the expansion of high-quality healthcare resources, achieving a balanced distribution, improving disease prevention and treatment at the grassroots level, and focusing on rural areas and communities. China's primary healthcare system is mainly composed of community health service centers and township health centers, with the former primarily serving urban residents and the latter mainly serving rural residents (1). With the implementation of hierarchical diagnoses and system pilot projects in cities and the continuous rapid growth of rural residents' healthcare needs with improved living standards, the healthcare service capacity of township health centers has significantly improved (2). According to the China Health Statistical Yearbook (3), as of 2021, China's rural township health centers had 2.84 beds per 1,000 people, 2.58 health technical personnel per 1,000 people, and 1.161 billion outpatient visits and inpatient admissions, with growth rates of 8.65, 6.02, and 2.37% per year, respectively, compared with the data from 2009. Both policies and data demonstrate that since the implementation

of the New Healthcare Reform, the healthcare service capacity of China's township health centers has made remarkable achievements (4), is undergoing a transition from "scale expansion" to "quality improvement," and is showing signs of advancing to a higher level. The reform strategy of "maintaining basic health services, strengthening primary healthcare, and establishing a mechanism" has been proposed since the New Healthcare Reform was implemented. Improving the primary healthcare service capacity is the best interpretation of "strengthening primary healthcare." However, China still faces problems such as inadequate primary healthcare service capacity (5), low primary healthcare service efficiency (6), and an uneven distribution of primary healthcare resources (7). Therefore, this study aims to explore the spatiotemporal evolution characteristics, leaping laws, and driving factors of the healthcare service capacity of township health centers in each province. By analyzing and presenting solutions for these issues, the results may assist policymakers in formulating macro-control policies that are suitable for China's healthcare system reform.

2 Literature review

Scholars at home and abroad have provided different definitions of healthcare service capacity based on three perspectives: resource, hierarchical, and functional. From the resource perspective, most scholars believe that human (8), financial (9), and material resources (10) constitute healthcare service capacity. From the hierarchical perspective, He and Yu (11) defined the primary healthcare service capacity using three levels: macro, meso, and micro, and asserted that primary healthcare service capacity is the resource allocation and operation mechanism of the primary healthcare service system, with the competence to continuously and effectively meet the primary healthcare service needs of the people. From the functional perspective, basic healthcare service capacity refers to the healthcare service capacity that primary healthcare centers can provide to the general public that is affordable and can meet their health needs (12).

Regarding the measurement of primary healthcare service capacity, scholars at home and abroad have mainly conducted empirical analyses from the efficiency perspective, and have frequently used data envelopment analysis (DEA). For example, Samut and Cafri (13) used DEA to evaluate hospital efficiency in 29 member countries of the Organization for Economic Cooperation and Development (OECD), and used efficiency as the dependent variable to analyze the impact of environmental factors on hospital efficiency using a tobit regression model. Ahmed (14) also used DEA to evaluate healthcare centers' management performance and technical efficiency. Yang (15) argues that although DEA is commonly used, the efficiency value of the decision-making unit (DUM) can only be 1, while the unexpected output super-efficiency slacks-based measure (SBM) model can allow the efficiency value of the decision-making unit to exceed 1, which is more applicable when measuring efficiency.

In recent years, research on spatiotemporal evolution has gradually increased and has been widely used in fields such as efficiency analysis (16), industrial structures (17), and resource environments (18). Han et al. (19) states that differential structures exhibit dynamic changes, including spatial and temporal attributes. Exploratory spatiotemporal data analysis methods are mainstream methods that combine spatial and temporal attributes for empirical analysis. Tim (20) proposes that these methods can help explain complex spatiotemporal relationships and can play a key role in multiple disciplines. Li and Zeng (21) used exploratory spatiotemporal data analysis methods to explore the spatiotemporal evolution characteristics of public health levels in Chinese cities, and found that these methods could more accurately depict healthcare centers' spatial distribution characteristics.

However, some deficiencies remain in research on the primary healthcare service capacity. First, regarding its efficiency analysis, most scholars have used the traditional DEA method. Although some scholars have used the non-expected output super efficiency SBM model to analyze the efficiency of healthcare services in China, it is only applied to the macro level measurement of healthcare service capacity efficiency and cannot explain the regional differences in grassroots healthcare service capacity (15, 22). Second, regarding spatiotemporal evolution, some scholars have used exploratory spatiotemporal data analysis methods (23); however, there is relatively little research on the driving factors of the spatiotemporal leap of primary healthcare service capacity using this method.

Therefore, this study uses the unexpected output super-efficiency SBM model to analyze the efficiency of the primary healthcare service capacity, with relevant data on township health centers in Chinese provinces from 2009 to 2021. It applies exploratory spatiotemporal data analysis methods to explore the heterogeneity and transition patterns of the primary healthcare service capacity, and uses quantile regression to analyze the driving factors of the spatiotemporal transition of the primary healthcare service capacity in each province. The results may provide a reference for policymakers to develop targeted policies that can enhance the healthcare service capacity of township health centers at the provincial level.

3 Materials and methods

3.1 Non-desirable output-super efficiency SBM model

In 2001, Tone (24) proposed the non-desirable output SBM (Super Efficiency SBM) model. This model differs from the traditional DEA model in that it addresses the issue of not considering slack variables due to the radial and angular problems between inputs and outputs. Tone later discovered that the model could not distinguish the results when all decision-making units were equal to 1. Therefore, he improved the SBM model and introduced the concept of super efficiency. To comprehensively measure the medical service capacity efficiency of township health

Abbreviations: SBM, Slacks-based measure; DUM, Decision-making units; lnpd, Rural population density; lnur, Urbanization rate; lnpcdi, Per capita disposable income; lnpGDP, Per capita gross domestic product; lnheex, Local governments' medical and health expenditure.

centers in various provinces in China, this study adopts the Non-Desirable Output-Super Efficiency SBM model, following Tone's calculation method. The specific formula is as follows:

$$\rho = min \frac{\frac{1}{m}\sum_{i=1}^{m}\frac{\bar{x}}{x_{il}}}{\frac{1}{S_1+S_2}\left(\sum_{q=1}^{S_1}\frac{y_q^g}{y_{ql}^g}+\sum_{r=1}^{S_2}\frac{y_r^g}{y_{rl}^g}\right)} \quad (1)$$

$$\begin{cases} \bar{x}_i \geq \sum_{j=1}^{n}\lambda_j x_{ij} \\ \bar{y}_q^g \leq \sum_{j=1}^{n}\lambda_j y_{qj}^l \\ \bar{y}_r^d \leq \sum_{j=1}^{n}\lambda_j y_{rj}^d \\ \bar{x}_i \geq x_{io};\ \bar{y}_q^w \leq y_{qo}^w;\ \bar{y}_r^b \geq y_{ro}^b \end{cases} \quad (2)$$

In this equation, ρ represents the medical service capacity efficiency value of township health centers in various provinces and cities. m is the number of input indicators, S1 is the number of desirable output indicators, S_2 is the number of non-desirable output indicators, and n is the number of decision-making units. $\bar{x}_i$, $\bar{y}_q^g$, $\bar{y}_r^d$ represent the quantities of input indicators, desirable output indicators, and non-desirable output indicators, respectively, considering slack variables.

3.2 Exploratory spatio-temporal data analysis method

Exploratory spatio-temporal data analysis method (25) (ESDA) is a combination of statistics and visualization. It constructs a spatial weight matrix to measure the spatial attribute correlation of neighboring areas and reveal the regional distribution pattern. The correlation is commonly represented by Moran's I index, which includes both global spatial autocorrelation and local spatial autocorrelation.

The formula for Global Moran's I, which represents the global spatial autocorrelation, is as follows:

$$GMI = \frac{n\sum_{i=1}^{n}\sum_{j=1}^{n}W_{ij}((x_i-\bar{x}_t)(x_j-\bar{x}_t))}{\sum_{i=1}^{n}\sum_{j=1}^{n}W_{ij}\sum_{i=1}^{n}(x_i-\bar{x}_t)^2} \quad (3)$$

In this equation, n represents the number of decision-making units, $\bar{x}_t$ t represents the average value of the medical service capacity efficiency for all samples at the grassroots level, x_i and x_j represent the values of neighboring provinces' spatial points, and, W_{ij} represents the spatial weight. Global Moran's I takes values between −1 and 1. When the Global Moran's I index is between −1 and 0, it indicates a negative correlation and a discrete pattern of medical service capacity efficiency among provinces. When the Global Moran's I index is 0, it indicates no correlation among provinces' medical service capacity efficiency. When the Global Moran's I index is between 0 and 1, it indicates a positive correlation and an agglomeration pattern of medical service capacity efficiency among provinces. The significance of the Global Moran's I index result is verified using the Z-value, for which the formula is as follows:

$$Z(GMI) = \frac{GMI - E(GMI)}{\sqrt{Var(GMI)}} \quad (4)$$

In this equation, $E(GMI)$ represents the theoretical expected value of GMI and $Var(GMI)$ represents the theoretical variance of GMI. When the Z-value passes the significance test, it indicates that the medical service capacity efficiency of township health centers in China exhibits positive spatial correlation.

Local spatial autocorrelation can analyze the correlation characteristics of medical service capacity efficiency among provinces' township health centers. Local spatial autocorrelation decomposes the Global Moran's I index to each province, obtaining the Local Moran's I index. The specific formula is as follows:

$$LMI = \frac{(x_i-\bar{x})\sum_{j=1}^{n}W_{ij}(x_j-\bar{x})}{\sum_{i=1}^{n}(x_i-\bar{x})^2} \quad (5)$$

In this equation, the symbols have the same meaning as Equation 4. The Local Moran's I index can partition each province into four quadrants: High-High (HH), Low-High (LH), Low-Low (LL), High-Low (HL). HH indicates that the medical service capacity efficiency of township health centers in the province itself and its neighboring provinces is high; LH indicates that the province itself has low values while its neighboring provinces have high values; LL indicates that the medical service capacity efficiency of township health centers in the province itself and its neighboring provinces is low; HL indicates that the province itself has high values while its neighboring provinces have low values.

Based on the spatial-temporal leap theory proposed by Rey and Janikas (26) and combined with the Moran's I scatter plot, four types of spatial-temporal transitions can be identified. Type I represents self-transition with stable surroundings, Type II represents self-stability with surrounding transitions, Type III represents transitions for both the province itself and its surroundings, and Type IV represents stability for both the province itself and its surroundings. The specific formula for the spatial stability of medical service capacity efficiency among provinces' township health centers is as follows:

$$S = \frac{F_{0,t}}{n} \quad (6)$$

In this equation, $F_{0,t}$ represents the number of sample provinces in period t that exhibit the "self-stability with stable surroundings" state of medical service capacity efficiency in township health centers. S value ranges from 0 to 1, where a larger S value indicates stronger spatial stability of medical service capacity efficiency and greater resistance to spatial transitions.

3.3 Quantile regression model

Quantile regression (27) is an econometric method proposed by Koenker and Bassett. Its characteristic is that it can split the dependent variable and estimate the trend of the independent variable's impact based on the splitting condition. The estimation results using weighted least absolute deviations are robust, not affected by outliers. The specific formula is as follows:

$$QR^{\theta} = \min X_i \sum_{i,LnY_I \geq X_i QR}^{\theta} |LnY_i - X_i QR| + \sum_{i,LnY_I X_i QR}^{1-\theta} |LnY_i - X_i QR| \quad (7)$$

TABLE 1 Indicators of efficiency and driving factors.

Indicator type	Indicator name	Indicator description
Input	Number of township health centers	Total number of township health centers in DUM (units)
	Number of healthcare professionals in township health centers	Total number of healthcare technicians in DUM (person)
	Number of beds in township health centers	Total number of beds in DUM (seats)
Expected output	Total number of diagnoses and treatments	Total number of consultations and treatments in township hospitals per year in DUM (number of people)
	Number of discharges	Total number of discharges from township hospitals per year in DUM (people)
Unexpected output	Average length of hospital stay	Average number of days spent hospitalized in township hospitals per year in DUM (day)
Driving factors	Population size	Rural population density (person/km^2)
	Regional development	Urbanization rate (%)
	Economic development	Per capita disposable income of rural residents (million yuan)
		Per capita gross domestic product (GDP; million yuan)

In this equation, QR^{θ} represents the estimated value at different quantiles, with θ representing the quantile. LnY_i represents the observed values at different quantiles, and X_iQR represents the predicted values at different quantiles. In this study, nine quantiles, namely 0.1, 0.2, 0.3, 0.4, 0.5, 0.6, 0.7, 0.8, and 0.9, are selected for analysis.

4 Indicator selection and data source

4.1 Indicator selection

This study selected the indicators by drawing on the previous research on the efficiency and driving factors of healthcare service capacity, and by considering data availability and other principles. To eliminate heteroscedasticity, this study logarithmically transformed the driving factors, as shown in Table 1.

This study's selected input indicators were the number of township health centers, the number of healthcare professionals in township health centers, and the number of beds in township health centers. Liu et al. (28) explains expected–unexpected output as follows: expected output can be understood as positive output that is determined by internal factors, while unexpected output concerns output that is better in smaller quantities. Researchers have shown that shortening the average length of hospital stay can improve the primary healthcare service capacity (29). Therefore, this study selected the total number of diagnoses and treatments and the number of discharges as the expected output indicators and the average length of hospital stay as the unexpected output indicator.

The driving factors were population size, economic development, policy support, and regional development. To measure population size, this study selected rural population density (lnpd) (30). Population density is a key indicator that reflects regional population distribution, while rural population density eliminates the problem of data untruthfulness caused by differences in the urban–rural population ratio. To measure regional development, this study selected the urbanization rate (lnur) (31). Urbanization refers to the process of the rural population moving to towns or cities as well as the process of transferring production factors to towns or cities. Researchers have shown that accelerated urbanization construction has a positive effect on narrowing the urban–rural gap and promoting urban–rural integration (32). To measure the economic development factors, this study selected rural residents' per capita disposable income (lnpcdi) (33) and per capita gross domestic product (GDP; lnpGDP) (34). Rural residents' disposable income reflects their consumption capacity from a micro perspective, while the per capita GDP can provide a more macro understanding of rural residents' living standards and is an important indicator for measuring macroeconomic development. To measure the policy support factor, this study selected the local governments' healthcare and health expenditure (lnheex) (35). Reasonable and long-term investment in healthcare and healthcare fiscal policies is considered a breakthrough concept for China to deepen its reform of the healthcare and healthcare system. Local governments' healthcare and health expenditure reflects their level of support for the healthcare and healthcare industries.

4.2 Data sources

Due to the absence of data for Beijing and Shanghai, this study focuses on the remaining 29 provinces. The relevant data for the years 2009-2021 were collected from the "China Health and Health Statistics Yearbook," "China Statistical Yearbook," and "China Rural Statistical Yearbook." China has been fully implementing the new healthcare reform since 2009, and selecting data with a long time span can also help eliminate the impact of short-term fluctuations on results, making analysis more stable and reliable.

TABLE 2 Efficiency values of the township health centers' healthcare service capacity by province.

Province	2009	2010	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	Mean	Rank
Nationwide	0.667	0.682	0.738	0.731	0.664	0.698	0.738	0.717	0.705	0.688	0.654	0.615	0.591	0.684	
Eastern	0.790	0.774	0.878	0.859	0.787	0.798	0.806	0.807	0.769	0.767	0.730	0.687	0.687	0.780	**2**
Tianjin	0.745	0.582	0.651	0.638	0.565	0.561	0.591	0.550	0.524	0.484	0.337	0.244	0.201	0.513	19
Hebei	0.560	0.557	0.599	0.567	0.519	0.539	0.581	0.624	0.557	0.541	0.461	0.419	0.361	0.530	18
Jiangsu	0.747	0.701	1.031	1.056	1.076	1.088	1.096	1.086	1.133	1.125	1.125	1.126	1.116	1.039	5
Zhejiang	1.221	1.240	1.284	1.300	1.290	1.296	1.303	1.301	1.309	1.317	1.312	1.328	1.330	1.295	1
Fujian	0.723	0.679	0.747	0.695	0.578	0.587	0.600	0.600	0.558	0.591	0.598	0.558	0.569	0.622	15
Shandong	0.680	0.727	1.041	1.080	0.742	0.768	0.747	0.779	0.808	0.792	0.728	0.731	0.777	0.800	13
Guangdong	1.121	1.160	1.088	1.053	1.057	1.048	1.032	1.028	0.814	0.860	0.880	0.820	0.901	0.989	6
Hainan	0.527	0.545	0.582	0.487	0.467	0.498	0.497	0.485	0.451	0.423	0.396	0.266	0.240	0.451	20
Central	0.658	0.707	0.783	0.817	0.691	0.781	0.866	0.876	0.897	0.894	0.828	0.802	0.752	0.796	**1**
Shanxi	0.307	0.301	0.351	0.342	0.307	0.309	0.314	0.331	0.359	0.315	0.278	0.251	0.196	0.305	26
Anhui	0.637	0.620	0.639	0.730	0.681	0.738	0.751	0.724	0.795	0.757	0.735	0.712	0.631	0.704	14
Jiangxi	1.031	1.022	1.058	1.006	0.738	0.798	1.019	1.019	1.030	1.016	0.774	0.680	0.671	0.912	10
Henan	0.788	1.011	1.018	1.018	0.936	1.037	1.066	1.089	1.082	1.116	1.089	1.123	1.134	1.039	4
Hubei	0.587	0.642	0.784	1.009	0.791	1.000	1.022	1.052	1.072	1.083	1.037	1.005	0.848	0.918	9
Hunan	0.599	0.646	0.846	0.797	0.691	0.803	1.023	1.041	1.046	1.078	1.054	1.042	1.033	0.900	11
Westward	0.659	0.679	0.714	0.692	0.652	0.682	0.726	0.666	0.643	0.619	0.609	0.573	0.545	0.651	**3**
Inner Mongolia	0.356	0.358	0.374	0.301	0.309	0.304	0.296	0.326	0.337	0.304	0.217	0.210	0.187	0.298	27
Guangxi	1.105	1.084	1.051	1.065	1.137	1.099	1.069	1.047	1.012	1.006	1.042	1.060	1.072	1.065	3
Chongqing	0.809	0.766	1.016	1.006	0.722	1.040	1.047	1.045	1.051	1.032	1.077	1.061	1.066	0.980	7
Sichuan	1.340	1.203	1.211	1.235	1.137	1.179	1.155	1.149	1.129	1.071	1.116	1.075	1.042	1.157	2
Guizhou	1.132	1.132	1.100	1.096	1.073	1.005	0.600	0.572	0.608	0.680	0.710	0.619	0.661	0.845	12
Yunnan	1.034	1.037	1.025	1.052	1.061	1.061	1.032	1.034	0.783	0.789	0.798	0.846	0.864	0.955	8
Xizang	0.257	0.324	0.305	0.272	0.277	0.259	0.249	0.306	0.302	0.259	0.181	0.131	0.087	0.247	28
Shaanxi	0.354	0.332	0.367	0.369	0.329	0.357	0.386	0.385	0.417	0.429	0.375	0.288	0.277	0.359	25
Gansu	0.124	0.401	0.455	0.455	0.412	0.397	0.423	0.471	0.507	0.505	0.455	0.420	0.362	0.414	21
Qinghai	0.398	0.436	0.539	0.431	0.387	0.375	0.324	0.377	0.361	0.306	0.286	0.268	0.251	0.365	23
Ningxia	0.557	0.592	0.568	0.492	0.454	0.490	1.362	0.484	0.488	0.428	0.402	0.371	0.337	0.540	17
Xinjiang	0.446	0.483	0.551	0.530	0.523	0.617	0.773	0.795	0.718	0.615	0.654	0.532	0.338	0.583	16
Northeast	0.389	0.398	0.371	0.377	0.328	0.330	0.344	0.368	0.395	0.346	0.287	0.215	0.192	0.334	**4**
Liaoning	0.391	0.399	0.452	0.452	0.400	0.411	0.417	0.437	0.473	0.442	0.375	0.281	0.234	0.397	22
Jilin	0.329	0.313	0.305	0.289	0.220	0.223	0.214	0.222	0.255	0.266	0.206	0.154	0.146	0.242	29
Heilongjiang	0.448	0.482	0.355	0.389	0.365	0.356	0.401	0.445	0.459	0.329	0.279	0.209	0.196	0.363	24

Bold text means regional ranking.

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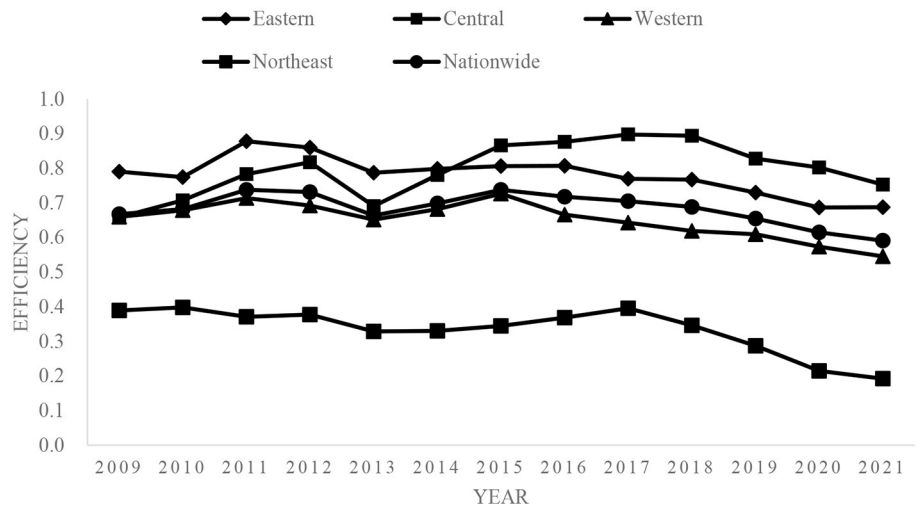

FIGURE 1
Evolution of provincial medical service capacity and efficiency.

TABLE 3 Global Moran's I values, 2009–2021.

Year	*I*	*E(I)*	*SD(I)*	*Z*	*P*
2009	0.348	−0.036	0.105	3.670	0.000
2010	0.386	−0.036	0.105	4.012	0.000
2011	0.413	−0.036	0.106	4.241	0.000
2012	0.400	−0.036	0.106	4.115	0.000
2013	0.311	−0.036	0.105	3.293	0.001
2014	0.327	−0.036	0.106	3.433	0.001
2015	0.272	−0.036	0.106	2.916	0.004
2016	0.355	−0.036	0.106	3.695	0.000
2017	0.349	−0.036	0.105	3.643	0.000
2018	0.390	−0.036	0.106	4.031	0.000
2019	0.355	−0.036	0.106	3.697	0.000
2020	0.355	−0.036	0.106	3.699	0.000
2021	0.379	−0.036	0.106	3.928	0.000

I, Moran's I value; E(I), Expected value; SD(I), Standard deviation; Z, Z value; P, P value.

5 Empirical result

5.1 Efficiency measurement result of healthcare service capacity

(1) Measurement result of the efficiency of the township health centers' healthcare service capacity.

This study used the SBM Run software to measure the efficiency of the healthcare service capacity of township health centers in 29 Chinese provinces from 2009 to 2021. The results are shown in Table 2.

(2) Result of the efficiency of the provincial township health centers' healthcare service capacity.

This study used Excel to create an efficiency evolution chart of the provincial healthcare service capacity in the eastern, central, western, and northeastern provincial, as well as nationwide, as shown in Figure 1.

5.2 Spatiotemporal evolution result of healthcare service capacity

(1) Empirical result of the global spatial autocorrelation

When the Global Moran's I value is between −1 and 0, the healthcare service capacity of township health centers in each province is negatively correlated and discrete; when the Global Moran's I value is 0, it indicates no correlation; and when the Global Moran's I value is between 0 and 1, it indicates a positive correlation in an agglomerated state. This study used the *z*-value to verify the significance of the Global Moran's I results. When the *z*-value passed the significance test, the healthcare service capacity of township health centers had a positive spatial correlation. This study used Stata 17.0 software to obtain the Global Moran's I values for the efficiency of the township health centers' healthcare service capacity in 29 Chinese provinces for 2009–2021. The results are shown in Table 3.

(2) Empirical result of the local spatial autocorrelation

Local spatial autocorrelation can be used to analyze the correlation characteristics of the healthcare service capacity of township health centers in each province. Local spatial autocorrelation decomposes the Global Moran's I of each province to obtain the Local Moran's I. The Local Moran's I divides each province into four quadrants: high-high (HH), low-high (LH), low-low (LL), and high-low (HL). HH represents a high value of healthcare service capacity of township health centers in the province and neighboring provinces, LH represents a low

TABLE 4 Quadrant distribution of the Local Moran's I, 2009–2021.

Year	First quadrant (HH)	Second quadrant (LH)	Third quadrant (LL)	Fourth quadrant (HL)
2009	Jiangsu/Zhejiang/Fujian/ Shandong/Guangdong/Jiangxi/ Henan/Guangxi/Chongqing/ Sichuan/ Yunnan/Guizhou	Hainan/Anhui/ Hubei/Hunan/ Qinghai/Xizang	Hebei/Shanxi/Inner Mongolia/Shaanxi/ Gansu/Ningxia/ Xinjiang/Liaoning/ Jilin/Heilongjiang	Tianjin
2015	Jiangsu/Zhejiang/ Shandong/Guangdong/ Jiangxi/Henan/Anhui/ Hubei/ Hunan/Guangxi/ Chongqing/Yunnan	Hainan/Fujian/ Qinghai/Xizang/ Guizhou	Hebei/Tianjin/Inner Mongolia/Shanxi/ Shaanxi/Gansu/ Liaoning/ Jilin/Heilongjiang	Sichuan/Ningxia/Xinjiang
2021	Jiangsu/Zhejiang/ Shandong/Guangdong/ Jiangxi/Henan/Anhui/ Hubei/Hunan/Guangxi/ Chongqing/Yunnan/ Guizhou	Hainan/Fujian/ Qinghai/Xizang	Hebei/Tianjin/ Inner Mongolia/ Shanxi/Shaanxi/ Gansu/Ningxia/ Xinjiang/Liaoning/ Jilin/Heilongjiang	Sichuan

value of healthcare service capacity in the province itself and a high value in neighboring provinces, LL represents a low value of healthcare service capacity in the province and neighboring provinces, and HL represents a high value of healthcare service capacity in the province itself and a low value in the neighboring provinces. Following Rey and Janikas (26) spatiotemporal leap theory combined with the Moran's I scatter plot, this study derived four spatiotemporal leap transition types: Type I, self-leaping and surrounding stability; Type II, self-stability and surrounding leaping; Type III, self-leaping and surrounding leaping; and Type IV, self-stability and surrounding stability. To better characterize the spatial agglomeration trend of each province, this study selected the Local Moran's I values of each province in 2009, 2015, and 2021, according to the efficiency trend changes. The results are shown in Table 4.

(3) Spatiotemporal leap result

This study analyzed the spatiotemporal leap pattern of the provincial township health centers' healthcare service capacity using the empirical local spatial autocorrelation results. The leap type results are shown in Table 5.

5.3 Result of the driving factors of healthcare service capacity

(1) Quantile regression result

This study used Stata 17.0 software to calculate the quantile regression values of the driving factors of healthcare service capacity. The results are shown in Table 6.

(2) Nested spatiotemporal leaps and quantile regression result

This study followed the quantile regression steps of Zhang et al. (36) and Ma et al. (37). This study used the 0.1–0.5 quantile as the low quantile response and the 0.6–0.9 quantile as the high quantile response. Based on the sign differences, the quantiles were divided into four pattern types: low quantile response and significant positive effect: low quantile-driven pattern; low quantile response and significant negative effect: low quantile-constrained pattern; high quantile response and significant positive effect: high quantile-driven pattern; and high quantile response and significant negative effect: high quantile-constrained pattern. The spatiotemporal leap results for 2009–2021 were nested within the four pattern types. The results are shown in Table 7.

6 Discussion

6.1 Efficiency analysis of medical service capacity

Regarding the changes in efficiency, the efficiency results for the healthcare service capacity of township health centers can be divided into two stages. The first stage is 2009–2015, during which the overall efficiency value shows an upward trend. The number of provinces with efficiency values greater than 1 increased from 7 in 2009 to 12 in 2015, with small fluctuations observed between 2011 and 2015. The policy research found that at this stage, healthcare service items were reformed because of township health centers' limited healthcare resources (38). Basic public health service items were mainly undertaken by township health centers, which weakened their healthcare service capacity to some extent. Subsequently, the Chinese government stimulated the development of township health centers through financial investment and other means. The second stage is 2016–2021, during which a downward trend in efficiency emerges. The number of provinces with efficiency values greater than 1 decreased from 11 in 2016 to 7 in 2021. During this period, the Chinese government implemented strict budget mechanisms from the administrative and economic perspectives, and township health centers gradually developed into nonprofit healthcare institutions. 2016 is the first year of the "13th Five-Year Plan". The Notice on Deepening the Key Tasks of the Reform of the Medical and Health System in 2016 triggered by The General Office of the State Council mentioned comprehensively deepening the reform of public hospitals. Put forward specific measures for the tasks of government responsibility, hospital management system and personnel system. From an economic point of view, the policy of canceling drug addiction has an impact on the income of township health centers, which means that township health centers are gradually developing into non-profit medical institutions. However, this approach also led to a reduction in the township health centers' profits (39). At the same time, since 2016, the number of township health institutions in most provinces

TABLE 5 Spatiotemporal leap types of the provincial township health centers' healthcare service capacity.

Leap type	2009–2015	2016–2021
Type I	HH-LH: Fujian/Guizhou LH-HH: Hunan/Hubei/Anhui HL-LL: Tianjin LL-HL: Ningxia/Xinjiang	LH-HH: Guizhou HL-LL: Ningxia/Xinjiang
Type II	HH-HL: Sichuan	-
Type III	-	-
Type IV	HH-HH: Jiangsu/Shandong/Chongqing/Henan/ Yunnan/Guangxi/Jiangxi/Guangdong/ Zhejiang LL-LL: Hebei/Shanxi/Shaanxi/Gansu/Liaoning/Jilin/Inner Mongolia/Heilongjiang	HH-HH: Jiangsu/Yunnan/Jiangxi/Guangxi/ Chongqing/Hunan/Guangdong/ Zhejiang/Henan/Hubei/Anhui/Shandong LH-LH: Fujian/Hainan/Qinghai/Xizang LL-LL: Gansu/Shanxi/Shaanxi/Tianjin/ Hebei/Jilin/Inner Mongolia/Liaoning/ Heilongjiang HL-H: Sichuan

TABLE 6 Results of quantile regression and ordinary least squares regression for the drivers of healthcare service capacity.

	con_s	lnpd	lnur	lnpcdi	lnpGDP	lnheex
Ordinary least squares	−2.394***	0.129***	−0.622***	−0.448**	0.136***	0.685***
0.1	−3.336***	0.130***	−0.868***	−0.040	0.113**	0.455
0.2	−2.382***	0.097***	−0.585***	−0.765	0.138***	0.941***
0.3	−1.984***	0.094***	−0.506***	−0.796**	0.144***	0.902***
0.4	−2.037***	0.117***	−0.495***	−0.914***	0.109***	1.05***
0.5	−1.884**	0.124***	−0.490***	−1.097***	0.133***	1.156***
0.6	−1.312	0.126***	−0.470***	−1.218***	0.196***	1.083***
0.7	−0.588***	0.126***	−0.434***	−0.765	0.222***	0.526
0.8	0.241***	0.126***	−0.262*	−0.564*	0.204***	0.231
0.9	−0.210	0.159***	−0.190	0.726	0.050	−0.597

***$p < 0.01$; **$p < 0.05$; * $p < 0.1$; *con_s: constant term.*

has decreased year by year, which is another reason for the change in efficiency value.

The provinces can be divided into five categories based on their average efficiency values. The first category is Zhejiang, Sichuan, Guangxi, Henan, and Jiangsu, with efficiency values in the range [1, ∞). The second category is Guangdong, Chongqing, Yunnan, Hubei, Jiangxi, and Hunan, with efficiency values in the range [0.9, 1), mainly in the central region. The third category is Guizhou, Shandong, Anhui, Fujian, Xinjiang, Ningxia, Hebei, and Tianjin, with efficiency values in the range [0.5, 0.9). The fourth category is Hainan, Gansu, Liaoning, Qinghai, Heilongjiang, Shanxi, and Shaanxi, with efficiency values in the range [0.3, 0.5). The fifth category is Inner Mongolia, Tibet, and Jilin, with efficiency values in the range [0, 0.3). The proportion of hierarchical diagnoses and treatment pilot projects in cities in the first category is relatively large; 11 prefecture-level cities in Zhejiang and 21 administrative units in Sichuan have become pilot cities for hierarchical diagnoses and treatments, and the efficiency values for both provinces remain above 1. This result is consistent with that of Gao et al. (40). Regarding the average annual growth rate, the rates for Gansu, Hunan, Jiangsu, Hubei, Henan, Chongqing, Shandong, and Zhejiang are positive, while those for the remaining 22 provinces are negative, which relates to the health funding allocated by local governments to township health centers.

Overall, the differences among the four major regions are quite obvious, with the general trend showing the order central > eastern > western > northeastern. This trend is consistent with Pei's results (41). Zhang (42) found that both the actual reimbursement rate for hospitalization and the number of residents who were hospitalized in township health centers were highest in the central region. Regarding the efficiency trend of healthcare service capacity in township health centers, the eastern region was the highest from 2009 to 2014 but was overtaken by the central region in 2015. The efficiency value began to decline in 2011 and showed a clear upward trend from 2021. In the central region, the efficiency value showed an upward trend for 2009–2012, a significant downward trend during 2012–2013, a rebound from 2013 to 2018, and a slight downward trend for 2018–2021. In the western region, the efficiency value shows a slight fluctuation trend from 2009 to 2013, displaying a reverse-U-shape, and shows an upward trend from 2013 to 2015, but continuously declined from 2015 to 2021. In the northeastern region, the efficiency value remained stable

TABLE 7 Nested results of spatiotemporal leaps and quantile regression.

Quantile response mode	Driving type	Leap province
Low quantile driving	Parallel development	LH-HH (Anhui/Hubei/ Hunan) LL-HH (0)
	Reverse development	LH-HL (0) LL-HL (0)
Low quantile constraining	Mutual constraint	LH-LH (Hainan/Qinghai/ Xizang) LL-LL (Hebei/Shanxi/ Shaanxi/Inner Mongolia/ Gansu/Ningxia/ Xinjiang/ Liaoning/Jilin/Heilongjiang)
	Reverse development	LH-LL (0) LL-LH (0)
High quantile driving	Parallel development	HH-HH (Jiangsu/Zhejiang/ Shandong/ Guangdong/Jiangxi/ Henan/Guangxi/ Chongqing/Yunnan/ Guizhou) HL-HL (0)
	Reverse development	HH-HL (Sichuan) HL-HH (0)
High quantile constraining	Mutual constraint	HH-LH (Fujian) HL-LL (Tianjin)
	Reverse development	HH-LL (0) HL-LH (0)

from 2009 to 2017 but showed a significant downward trend from 2017 to 2021. Thus, the efficiency measurements of the healthcare service capacity of township health centers in the four regions declined at different times. In the interviews conducted by the research team in 2018 in Shandong, Zhejiang, Gansu, and other places, the respondents revealed that rural residents' increasing health demands could not be met by the healthcare resources and technical skills held by township health centers. For example, the township health centers could only perform simple auxiliary examinations, such as X-rays and ultrasounds, while routine auxiliary examinations, such as CT scans, could only be performed in county-level hospitals or higher. Moreover, regarding the per capita healthcare expenses, the proportion of personal expenditure has continued to rise, which has led rural residents to be more inclined to choose county- or city-level hospitals for healthcare treatment.

In conclusion, the efficiency of healthcare service capacity in township health centers is better in provinces with a higher proportion of hierarchical diagnoses and treatment pilot projects in cities. In terms of regional distribution, the efficiency of healthcare service capacity generally shows the order of "central > eastern > western > northeastern," and the efficiency of the healthcare service capacity in township health centers in the four regions has declined at different times. China should expand the pilot program of tiered diagnosis and treatment in cities. Provinces should fully utilize administrative measures and economic levers to form an incentive mechanism for preliminary diagnoses at the grassroots level, two-way referrals, and multichannel guidance; promote the expansion and sinking of high-quality healthcare resources; alleviate the structural imbalance of healthcare resource allocation; and improve the grassroots healthcare service capacity by accelerating the construction of national and regional healthcare centers, expanding the construction of healthcare consortia, and improving relevant policies for tiered diagnoses and treatments.

6.2 Space-time evolution analysis of medical service ability

The Global Moran's I values for the efficiency of the healthcare service capacity in each year are positive and pass the test at different levels of significance, indicating that the efficiency of the healthcare service capacity has a significant spatial correlation with the spatiotemporal distribution. The Global Moran's I results can be divided into three stages. The first stage is 2009–2011, during which the efficiency of the healthcare service capacity exhibits a continuous upward trend. The second stage is 2011–2015, during which the efficiency of the healthcare service capacity shows an overall downward trend, except for a slight increase from 2013 to 2014. The third stage is 2015–2021, during which the efficiency of the healthcare service capacity shows a fluctuating upward trend. The efficiency trend changes in the healthcare service capacity are increasingly influenced by neighboring provinces, and there is both competition and cooperation among the provinces. Accordingly, China has implemented two assistance mechanisms: vertical and horizontal. The vertical assistance mechanism refers to the support between healthcare consortia, whereas the horizontal assistance mechanism refers to the support between healthcare centers at the same level. Township health centers have also developed network-style layouts.

From an overall perspective, over the period 2009–2021, the efficiency of the healthcare service capacity in each province is mainly distributed in the HH and LL quadrants, indicating a strong spatial clustering pattern with predominantly positive spatial autocorrelation. Regarding the provincial changes, in 2009, 12 provinces are in the HH quadrant and 10 are in the LL quadrant. In 2015, 12 provinces are in the HH quadrant and 9 provinces are in the LL quadrant. In 2021, 13 provinces are in the HH quadrant and 11 are in the LL quadrant. The provincial trend further confirms that the spatial distribution of the healthcare service capacity has a strong positive correlation, with relatively few provinces in the LH and HL quadrants, thus exhibiting a "high agglomeration, low differentiation" spatial distribution.

Regarding the regional trend, in 2009, the HH agglomeration pattern is mainly distributed in the eastern region. The relevant GDP data shows that the eastern region's economic development is faster than that of other regions, and that more healthcare resources can be supplied to a certain extent, which is conducive to improving the healthcare service capacity of township health clinics. The LL agglomeration pattern is mainly distributed in the western and northeastern regions, where there are relatively more rural populations and a greater consumption of primary healthcare resources. The central region mainly exhibits an LH agglomeration pattern, indicating that the positive influence of neighboring provinces increases the efficiency of the healthcare service capacity. In 2015, the HH agglomeration pattern starts to spread to the central region, as backup advantages [Human, Financial, and Material resources (43)] are utilized and the corresponding investments are increased, resulting in faster

efficiency improvement. The LL agglomeration pattern mainly occurs in the western and northeastern regions. In 2021, the differences are smaller than in 2015, and the western and northeastern regions mainly exhibit the LL agglomeration pattern, indicating that China should focus on these regions.

The provinces mostly show the Type IV leap pattern, while no province shows the Type III pattern, indicating that the overall spatiotemporal evolution of each province is stable, and there are no features of extreme instability. Specifically, from 2009 to 2015, Fujian and Guizhou have a leap type of HH-LH. Fujian is a coastal province with rapid economic development and increasing demand for healthcare services, whereas Guizhou is affected by its neighboring provinces, leading to a decrease in the efficiency of healthcare service capacity. Hunan, Hubei, and Anhui were positively influenced by their neighboring provinces, leaping from LH to HH in terms of healthcare service capacity, while Tianjin's healthcare service capacity declined due to its negative neighboring influence, leaping from HL to LL. Ningxia and Xinjiang have a leap type of LL-HL, which may relate to the greater supply of healthcare resources due to economic development. Sichuan shows a positive local influence, resulting in a decrease in the healthcare service capacity of its neighboring provinces, and leaps from HH to HL. From 2016 to 2021, Guizhou showed a leap type of LH-HH due to a positive neighboring influence, leading to an increase in its healthcare service capacity, whereas Ningxia and Xinjiang are negatively influenced by neighboring provinces, leading to a decrease in their healthcare service capacity, with both provinces having a leap type of HL-LL.

In conclusion, the global spatial autocorrelation results indicate that the efficiency of healthcare service capacity in township health centers in each province has a significant spatial correlation with its spatiotemporal distribution, gradually forming a grid layout. From the local spatial autocorrelation results, the efficiency of healthcare service capacity mainly reveals the HH and LL distribution patterns, showing the characteristics of "high agglomeration, low differentiation." From the spatiotemporal leap results, the provinces mostly show the Type IV leap pattern, while no province shows the Type III pattern. China should establish national collaborative development models. Provincial governments should adopt a model of deep cooperation; build a nationwide healthcare information system that is interconnected and shared; and improve support mechanisms, such as multi-point practice and counterpart support regarding systems and technologies. They should also promote the excellent experiences and practices of township health centers that have good development status, provide a reference for township health centers northeastern, and promote the innovative development of township health centers based on their previous experience and actual local conditions, so as to effectively improve the grassroots healthcare service capacity.

6.3 Analysis of influencing factors of medical service ability

Lnpd was significantly tested at each quantile (0.1–0.9); the results showed a promoting effect on the efficiency of healthcare service capacity. This is because areas with denser rural populations require more healthcare resources, which stimulates township health centers' healthcare service capacity to some extent. Moreover, in reality, areas with high rural population density tend to receive a certain proportion of increased government investment, which is consistent with the results of Chen and Han (44). Lnur shows a statistical difference in all quantiles except the 0.9 quantile, indicating an inhibitory effect. Although urbanization drives rural development, it also leads to the flow of rural populations to urban areas, resulting in some rural areas becoming empty-nest villages. This causes both a healthcare concentration and a decline in the efficiency of the healthcare service capacity of township health centers; this result is consistent with that of Wang and Han (45). Lnpcdi is significantly tested in the low (0.3–0.5) and high (0.6, 0.8) quantiles; the coefficients were negative, indicating that the per capita disposable income of rural residents has an inhibitory effect on the efficiency of healthcare service capacity. This may be because as rural residents' income increases, their demand for healthcare becomes more urgent. They believe that urban hospitals have more abundant healthcare resources and are better able to meet their health needs, resulting in a decline in the efficiency of healthcare service capacity of township health centers, which is consistent with the results of Yang and Lü (15). LnpGDP is not significant in the 0.9 quantile, the other quantiles show statistical differences and exhibit a promoting effect. Generally, the higher a province's economic development level, the more funds the government can disperse for grassroots healthcare services. Jin et al.'s (46) results also indicate that in the New Healthcare Reform stage, regions with better economic development have smaller differences in the supply of basic healthcare resources and decreases in the differences in per capita GDP.Lnheex is significant in the low quantile (0.2–0.5) and high quantile (0.6) tests, and the coefficients are positive, indicating that it has a promoting effect on the efficiency of healthcare service capacity of township health centers. Increasing local governments' healthcare and health expenditure positively affects reducing residents' healthcare expense burden. According to the studies, from the supply perspective, local governments' healthcare and health expenditure is mainly used for healthcare infrastructure and healthcare staff salaries. Having fully-equipped healthcare infrastructure in township health centers can effectively improve rural residents' healthcare needs, while an increase in healthcare staff salaries can effectively improve the quality of healthcare services provided. However, from the regression results, local governments' healthcare and health expenditure reaches saturation after a certain increase, which is similar to the result of Wang and Hu (47).

In the low quantile-driving pattern, lnpd, lnpGDP, and lnheex are the most significant factors that affect the spatiotemporal efficiency leap of the healthcare service capacity of township health centers. They have a significant promoting effect on the development of the healthcare service capacity in Anhui, Hubei, and Hunan. In the low quantile-constraining pattern, lnur and lnpcdi constrain the spatiotemporal leap in the healthcare service capacity of township health centers. In this mode, the rapid promotion of urbanization and improvement of per capita disposable income is conducive to enhancing the healthcare service

capacity of township health centers in Hebei, Shanxi, and Inner Mongolia. In the high quantile-driving pattern, lnpd, lnpGDP, and lnheex have a significant promoting effect on the spatiotemporal leap in the healthcare service capacity of township health centers. This pattern is mainly observed in the eastern region, indicating that the economic development characteristics of provinces such as Jiangsu, Zhejiang, and Shandong promote the enhancement of the healthcare service capacity of township health centers. Local governments' fiscal health expenditure passes the significance test in the high quantile response (0.6), indicating that it will have a significant promoting effect when it reaches a certain level. In the high quantile-constraining pattern, lnur and lnpcdi are the main factors that constrain the spatiotemporal leap in the healthcare service capacity of township health centers. With the continuous acceleration of urbanization and improvement of residents' living standards, provinces such as Fujian and Tianjin constrain the spatiotemporal leap in the healthcare service capacity of township health centers. Judging from the characteristics of these two provinces, the urbanization level is relatively high, and the rural residents are gradually moving to the cities. Therefore, the development of township health centers will inevitably be affected to some extent, which is consistent with the actual situation.

In conclusion, the quantile regression results show that rural population density and per capita GDP significantly promote the improvement of the healthcare service capacity, and local governments' healthcare and health expenditure has a promoting effect on the healthcare service capacity of township health centers in certain quantiles. The urbanization rate and per capita disposable income inhibit the improvement of the healthcare service capacity of township health centers at certain quantiles. From the nested results of the spatiotemporal leaps and quantile regression, rural population density and local governments' healthcare and health expenditure promote the healthcare service capacity of township health centers in Anhui, Hubei, Hunan, and other provinces. Per capita GDP promotes the healthcare service capacity of township health centers in Jiangsu, Zhejiang, Shandong, Guangdong, and other provinces, whereas the level of urbanization restricts the leap in healthcare service capacity of township health centers in Tianjin and Fujian. China should guide each province in formulating implementation plans that meet their actual needs. Provinces such as Jiangsu, Zhejiang, Shandong, and Guangdong can adopt differentiated measures to improve their healthcare service capacity, such as by increasing local per capita income and increasing investment in healthcare and healthcare. Doing so will also have a radial effect on the neighboring provinces. Tianjin and Fujian Provinces can improve their grassroots healthcare service capacity by adjusting for population size and other measures.

7 Conclusion

Through the analysis of the medical and health service capacity of township hospitals in China, the efficiency of graded diagnosis and treatment pilot areas is higher. But in terms of time, efficiency has declined in all regions.The characteristics of "high aggregation, low differentiation" indicate that the ability of medical and health services will be affected by neighboring provinces. In addition, due to the differences in population, economy and other factors, the shortcomings of the medical service capacity of township health centers in various provinces are also different. The state should expand the trial of graded diagnosis and treatment in cities, establish national collaborative development models, and guide each province in formulating implementation plans that meet their actual needs.

8 Innovation and deficiency

The purpose of this paper is to accurately grasp the regional differences in the development of medical service capacity of township health centers in China, and to improve the medical service capacity of township health centers is a key issue for the government to actively promote the reform of the medical and health system. This paper comprehensively uses the non-expected output SBM model, exploratory spatiotemporal data analysis method and quantile regression to study the efficiency analysis, spatiotemporal evolution characteristics and driving factors of the medical service capacity of township health centers in China, which can fully reflect the quality of the medical service capacity of township health centers in China, and has a guiding role in regulating the medical service of township health centers. It can also be used for reference in other areas.

Although this study systematically and empirically assessed the efficiency, spatiotemporal evolution characteristics, and driving factors of the healthcare service capacity of township health centers in Chinese provinces, the data on the township health centers at the provincial level only allowed for the characterization of strategies for improving the healthcare service capacity of township health centers from a top-down perspective. However, there may be significant differences in the specific situation of each township in each province, which could lead to deficiencies in the institutional construction and bottom-up logic. Therefore, future research should collect and study the micro data on each township's health center.

Further, the driving factors selected herein were only obtained through a literature review, which may give rise to endogeneity issues owing to the lack of identification of critical factors in practical processes. Therefore, future research should conduct in-depth interviews to further explore the factors that may affect the healthcare service capacity of township health centers.

Data availability statement

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found at: https://data.stats.gov.cn/easyquery.htm?cn=C01 and http://www.nhc.gov.cn/mohwsbwstjxxzx/tjzxtjcbw/tjsj_list.shtml.

Author contributions

Among the contributing authors of this study, HC was engaged in study design, data processing, and analysis and paper writing. LZ was engaged in study design and involved in data analysis. JY actively participated in study design and field survey. All authors of this article have read and approved the final manuscript.

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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.

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Describing financial toxicity among cancer patients in different income countries: a systematic review and meta-analysis

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Background: There is limited evidence of financial toxicity (FT) among cancer patients from countries of various income levels. Hence, this study aimed to determine the prevalence of objective and subjective FT and their measurements in relation to cancer treatment.

Methods: PubMed, Science Direct, Scopus, and CINAHL databases were searched to find studies that examined FT. There was no limit on the design or setting of the study. Random-effects meta-analysis was utilized to obtain the pooled prevalence of objective FT.

Results: Out of 244 identified studies during the initial screening, only 64 studies were included in this review. The catastrophic health expenditure (CHE) method was often used in the included studies to determine the objective FT. The pooled prevalence of CHE was 47% (95% CI: 24.0–70.0) in middle- and high-income countries, and the highest percentage was noted in low-income countries (74.4%). A total of 30 studies focused on subjective FT, of which 9 used the Comprehensive Score for FT (COST) tool and reported median scores ranging between 17.0 and 31.9.

Conclusion: This study shows that cancer patients from various income-group countries experienced a significant financial burden during their treatment. It is imperative to conduct further studies on interventions and policies that can lower FT caused by cancer treatment.

KEYWORDS

direct medical cost, direct non-medical cost, indirect medical cost, catastrophic health expenditure, perceived financial hardship, systematic review, meta-analysis

1 Introduction

Cancer is the leading cause of death worldwide (1). Cancer cases and deaths continue to increase worldwide in both developed and developing countries. Despite the high cancer incidence rate in developed countries, the mortality rate is higher in developing countries (2, 3). Every year, approximately 400,000 youngsters are diagnosed with cancer (4). The growing number of people diagnosed with cancer places a responsibility on governments to offer services that are suitable, easily accessible, and reasonably priced. However, high-quality services for preventing, detecting, diagnosing, treating, supporting, and caring for those who have survived cancer are challenging to achieve due to multiple influential factors, including unstable politics, inadequately trained cancer care providers, and deficient coordination, in addition to the rising costs associated with cancer treatment (5).

A significant obstacle preventing many cancer patients from receiving therapy and care is the expense of doing so, given the significant geographical variations in patients' financial capabilities and preparedness to spend money on healthcare and wellness services (6). In the vast majority of low-resourced countries, there is either very little or no universal access insurance coverage for medical care. However, even among insured patients, a significant number are not adequately protected against the expensive requirements of cancer treatment due to the elevated costs of insurance, which include higher co-payments and rising deductibles. Hence, cancer patients typically must pay a significant portion of their treatment costs out of pocket (7). The medical and non-medical expenses of cancer care, which result in a financial burden for cancer patients, are not adequately described in the existing body of research due to the absence of a nomenclature that is consistent throughout the field. Recent research has led to the development of a comprehensive definition of FT, which may be summarized as "The possible consequence of perceived subjective financial distress caused by an objective financial burden" (8). The terms "direct costs" and "indirect care-related costs" refer to "objective financial burden," but "subjective financial hardship" refers to "material, psychosocial stress, negative feelings, and behavioral reactions to cancer care" (6, 8). FT is a term that is sometimes used interchangeably with terms such as financial or economic difficulties, financial difficulty, financial risk, and economic stress (9). Several studies provided valuable insights into the issue of FT among cancer patients and survivors. Yousuf Zafar (2016) highlighted that FT is a complex problem that affects the quality of life of cancer patients and survivors (10). Tucker-Seeley et al. (2016) build on this, indicating that socioeconomic factors contribute to FT, exacerbating health disparities (11). Arastu et al. (2020) highlighted that financial toxicity can be more prevalent among older adults, and they call for age-appropriate interventions (12). Gordon et al. (2016) found that cancer survivors face additional economic difficulties, such as out-of-pocket expenses and lost of income (13). Baddour et al. (2021) examined the objective and subjective impacts of financial toxicity on head and neck cancer survivors. They emphasize that financial distress not only affects the ability to pay for healthcare but also affects one's mental health and wellbeing (14).

Ramsey et al. (2013) highlighted that cancer patients are at a greater risk of bankruptcy than individuals without cancer and that the problem of FT can persist beyond cancer diagnosis and treatment (15). Ramsey et al. (2016) further highlighted the connection between FT and early mortality among cancer patients, as financial distress may result in reduced adherence to medical treatments and a lower overall survival rate (16).

In summary, FT is a complex and significant issue for cancer patients and survivors, and its negative consequences can be long-lasting. The studies listed here provide evidence of the widespread occurrence of FT among cancer patients and survivors, indicating the need for policies and interventions to mitigate its effects and improve the quality of life for those affected.

Our study aimed to bridge the current gap in knowledge on FT among cancer patients across different countries with varying income levels. Although recent systematic reviews have examined FT in either low- or high-income countries (17, 18), there is limited comprehensive evidence that explores FT among cancer patients globally.

To address this gap, this systematic review and meta-analysis of existing literature aim to determine the prevalence and measurement of both objective and subjective FT among cancer patients. Our study utilized a rigorous methodology to identify and evaluate relevant studies from diverse sources and synthesize their findings. Through this approach, we aimed to provide a comprehensive understanding of the financial burden that cancer patients face and how it affects their lives.

The current study aims to contribute to the existing knowledge of FT among cancer patients, which will help improve clinical practice and healthcare policies worldwide. Our study will also provide a platform for future research in this area, as we anticipate identifying areas where more research is needed. Ultimately, our findings will assist in addressing the needs of cancer patients and survivors and support the development of effective interventions to mitigate the negative impacts of FT.

2 Methods

2.1 Search strategy

The systematic review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (19) guidelines from 15 August 2022 to 15 January 2023. The protocol was registered in Open Science (Registration DOI: https://doi.org/10.17605/OSF.IO/MUNKG; Supplementary Table S1: PRISMA checklist).

PubMed, Science Direct, Scopus, and CINAHL databases were searched to find articles that reported FT. The search was built based on the research question concerning population/problem (cancer), outcome (financial toxicity), and exposure (healthcare treatment) and (cost of illness), as well as their synonyms (Supplementary File S1: search strategy). There was no limit on the publication year, design, or setting of the study, in order to minimize underreporting bias. In addition, a manual search through the reference list of eligible studies was applied. The search hits for databases are provided in the Supplementary File.

The primary outcome was to find the prevalence of subjective and objective FT among cancer patients. The cost of treatment was also considered a secondary outcome.

2.2 Study selection

First, the authors formed a search strategy involving all the relevant keywords based on their knowledge and literature. All search results were transferred to the Endnote X9 software. A total of 244 articles were identified through an online search and 24 articles through a manual search. Then, the duplicate articles were eliminated (20). The titles and abstracts of the remaining 185 articles were screened by two independent reviewers (MMA and VK). Subsequently, a total of 53 articles were retained for full-text review. Disagreements between the two reviewers were resolved by involving a third author (WMA). After a full-text review of the 53 articles, 40 were selected using the on-line search and 24 articles were selected using a manual search. The eligibility of the included articles was agreed upon by all authors. The PRISMA flowchart demonstrated the screening process (Figure 1).

We included studies that are original English quantitative research articles and reported the financial toxicity (objective and subjective) of any type of cancer that were published before 28 August 2022. In addition, studies that reported any cost of cancer, including direct medical, direct non-medical, and indirect costs, were included. This study did not assess the intangible cost as it is difficult to calculate its monetary value. Economic evaluation studies, conference abstracts, reviews, and qualitative studies were excluded. The reasons for these exclusions are as follows: the economic evaluation studies might include the cost of cancer but do not address the primary aim of this review, namely, FT. Conference abstracts do not always present consistent and dependent data. Reviews were excluded because the designed protocols were different from this study; in addition, the outcome evaluation methods were different. Qualitative studies are more of a subjective nature, which cannot be pooled as per the protocol, and their analysis is different from the quantitative data.

2.3 Data extraction and quality assessment

The data were presented based on author date, type of cancer, study participants (sample size and sociodemographic characteristics such as age and gender), the prevalence of FT (subjective and objective), cost of illness, tools used to measure the FT, and quality scoring (Supplementary Table S2). The cost of illness is classified into direct and indirect costs. Direct costs are expenses that can be directly and specifically traced to a specific cost object (for example, the medicines consumed by a patient during his/her hospital stay). In contrast, indirect costs are defined as "expenses that cannot be directly linked to a specific cost object (e.g., labour costs) (21). The quality of all included articles was assessed using the *Newcastle – Ottawa quality assessment scale* for cohort and cross-sectional studies (adapted for *cross-sectional studies*), which comprises three sections: selection, comfortability, and outcome. The quality score is shown in Supplementary Table S2.

2.4 Data synthesis and analysis

Quantitative data were used to find the prevalence of FT. Review Manager 5.3 software was utilized to run the meta-analysis of quantitative data-reported studies. A random-effects meta-analysis was used to calculate pooled data with 95% confidence intervals (CIs). The I^2 index was utilized to assess the heterogeneity among studies, with values classified of ≤25%, 26–50%, and >50% as low, moderate, and high heterogeneity, respectively (22, 23).

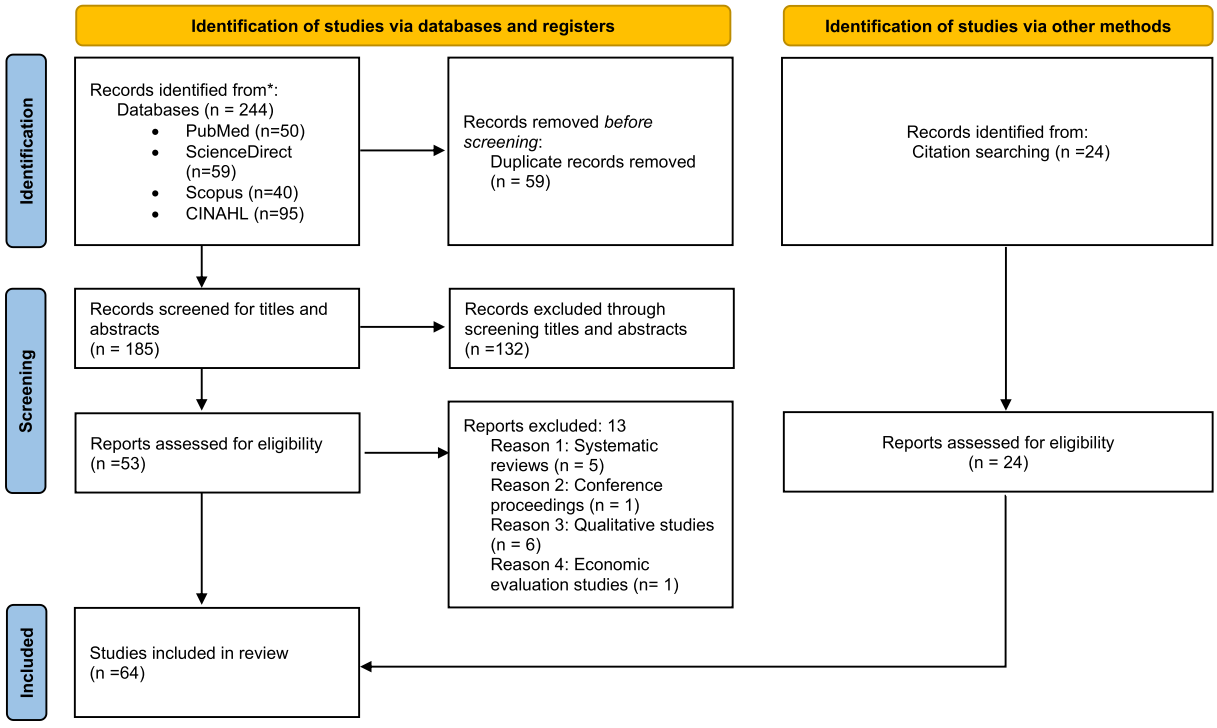

FIGURE 1
Flowchart of literature review search per Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA 2020) guidelines.

3 Results

3.1 Description of studies

The included studies were carried out worldwide, including in the USA ($n = 30$) (24–53), Europe ($n = 10$) (20, 54–62), Canada ($n = 4$) (63–66), Malaysia ($n = 4$) (67–70), Australia ($n = 6$) (71–76), Brazil ($n = 2$) (77, 78), China ($n = 2$) (79, 80), Iran ($n = 2$) (81, 82), Korea ($n = 1$) (83), Taiwan ($n = 1$) (84), Japan ($n = 1$) (85), and Ethiopia ($n = 1$) (86). A total of 47,964,650 cancer patients participated in a total of 64 studies carried out worldwide, with study samples ranging from 26 to 19.6 million. Out of the 64 studies, 15 studies included participants with any type of cancer (32, 35, 36, 39, 41, 44, 51, 52, 58, 65, 68, 72, 84–86), a mix of 2 or more types of cancers (10 studies) (27, 33, 38, 53, 56, 62, 69, 71, 79, 81), breast cancer (11 studies) (30, 40, 45–47, 50, 55, 66, 76, 82, 83), colorectal cancer (6 studies) (48, 57, 59, 67, 70, 75), colon cancer ($n = 1$) (49), skin cancer (3 studies) (20, 77, 78), lung cancer (2 studies) (54, 80), lung cancer with brain metastasis (1 study) (34), prostate cancer (3 studies) (63, 64, 74), pancreatic cancer (1 study) (28), bladder cancer (2 studies) (29, 31), head and neck cancers ($n = 3$ studies) (43, 60, 61), blood cancer (3 studies) (24, 26, 73), liver cancer (1 study) (42), gynecologic cancer (1 study) (25), and multiple myeloma ($n = 1$) (37).

3.2 Measurement of objective financial toxicity

Included research is rarely concentrated, particularly on measurable indicators of FT. Only five studies provided the measurement of objective FT in terms of the prevalence of catastrophic health expenditure (CHE), which was defined as a healthcare cost-to-income ratio of more than 40% in four studies (69, 70, 79, 80) and as the out-of-pocket payment (OOP) that exceeds 10% of total household income in one study (86).

3.3 The pooled prevalence of objective financial toxicity

A study conducted in Ethiopia reported a 74.4% prevalence of CHE. Two studies were carried out in Malaysia, one among colorectal cancer patients and the other among prostate, bladder, and renal cancer patients, and 47.8 and 16.1% of respondents, respectively, reported having experienced CHE (70, 80). Two studies were carried out in China; one study showed a total of 72.7% of participants experienced catastrophic health spending (69), and the other one showed the prevalence according to the state, where it was 87.3, 66.0, 33.7, and 19.6% in Chongqing, Fuzhou, Beijing, and Shanghai states, respectively (79). We pooled the findings of the last study (79) before including them in the meta-analysis; therefore, the prevalence of CHE in Mao et al. (79) was 51.65%. The last four studies enabled meta-analysis as they used the same method of measuring the CHE. As such, the pooled prevalence of CHE was 0.47 (95% CIs: 0.24–0.70), and the heterogeneity was high ($I^2 = 99\%$) (Figure 2).

3.4 Measurement of subjective financial toxicity

In total, 30 studies provided data on subjective FT (24–27, 29, 31, 32, 37, 39, 41, 44–46, 49–52, 58, 59, 61, 62, 64, 67–69, 71, 73, 75, 80, 85). The measures of FT varied widely among the studies. Nine of them used the COmprehensive Score for financial Toxicity (COST) tool, which had 11 items and a score ranged from 0 to 44, where lower COST values indicating higher financial toxicity (25, 26, 31, 37, 46, 68, 71, 73, 85). The median COST score in the included studies ranged between 17 and 31.9. The lowest score was reported among patients with acute myeloid leukemia and the highest among patients with gynecological cancers. Moreover, 10 studies used a 4- to 7-point Likert scale to assess the prevalence of subjective FT (24, 29, 45, 58, 59, 61, 62, 64, 67, 80), where the reported prevalence ranged between 20.9 and 83.7%. In addition, two studies used the median of COST as a cutoff point to assess those with and without FT (25, 26). One study used the Personal Financial Well-Being Scale (PFLBS), which consisted eight items on a Likert scale of 10 points, where 1–4 indicated high FT and LWB, 4.1–6.9 indicated average FT, and 7–10 indicated low FT and high financial WL (69). In addition, one study used four questions with "yes and no" answers to assess the subjective FT; those who responded "yes" to at least one of the four questions were defined as experiencing financial toxicity (44). Ekwueme et al. (2019) described the FT as material hardship and psychological hardship and found it to be 25.3% and 34.3% among the study participants, respectively. Three studies used four questions related to debt incurred, worry about paying bills, and making financial sacrifices as a measure of FT (27, 41, 49). In addition, one study in Australia assessed the FT using three questions related to perceived prosperity, financial strain, and the ability to raise money in an emergency (75) (Table 1).

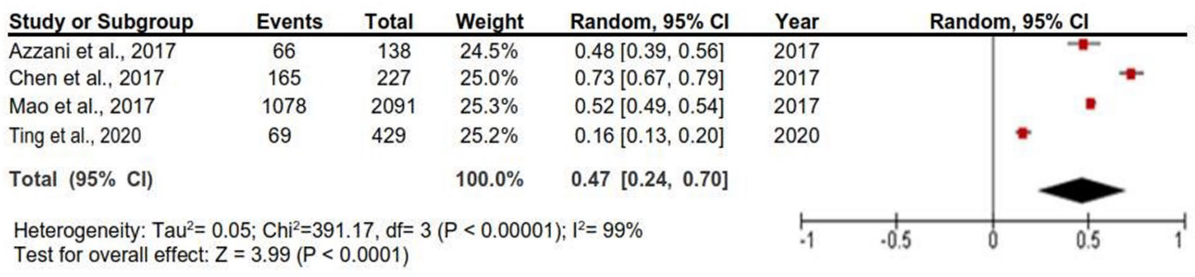

FIGURE 2
Random-effects meta-analysis of studies that reported the prevalence of catastrophic health expenditure.

TABLE 1 Prevalence of subjective financial toxicity in included studies.

Author, Year	Country	Cancer type	Year of research	Sample size	Prevalence of FT (%)	Tools used
El-Haouly et al. (2020) (64)	Canada	Prostate cancer	2020	171	22.3%	6-point Likert scale ("not a burden," "light burden," "moderate burden," "considerable burden, but sustainable," "considerable burden that is difficult to manage and stressful," and "unsustainable burden"). A categorization was achieved according to the reporting of a moderate/considerable/unsustainable burden (yes/no).
Azzani et al. 2016 (67)	Malaysia	Colorectal cancer	2016	138	20.9%	5-point Likert scale ('very difficult', 'difficult', 'somewhat difficult', 'not that difficult', or 'not difficult at all'). A categorization was achieved according to the reporting of a difficult/very difficult (yes/no).
Chen et al. 2017 (80)	China	Lung cancer	2017	227	83.7%	5-point Likert scale [somewhat, quite a bit, and very much (toxic group) versus not at all or a little bit (non-toxic group) from the COST-PROM questionnaire].
Perry et al. 2019 (45)	USA	Breast cancer	2011–2017	309	37.5% (F strain) 26.1% (FT)	Financial strain is assessed by a four-item checklist that asks participants to indicate whether their income is sufficient to allow them to afford: (1) food and housing, (2) clothing, medicine, and home repairs, (3) going out for a meal and entertainment, and/or (4) a week-long vacation, health permitting. Participants were classified as financially strained if they indicated that they could not afford one or more of the four options. FT was assessed by a 5-point Likert scale: strongly disagree (1) to strongly agree (5), agree and strongly agree are considered as having FT.
Pearce et al. 2018 (58)	Europe	All cancer type	2009–2015	2,931	22%	4-point Likert scale from EORTC QLQ-c30, a little, quite a bit, very much as having FT, not at all as no FT.
Sharp et al. 2018 (59)	Europe	Colorectal cancer	2007–2009	493	41% had financial stress, 39% financial strain, 32% reported both financial stress and financial strain	7-point Likert scale: more difficulty more concern to much less concern, collapse into more difficulty/concern, no change, less difficulty/concern. Financial stress was assessed as the impact of the cancer diagnosis on the household's ability to make ends meet, and financial strain was assessed as the impact on the individual (i.e., how the respondent had felt about their household's financial situation since their cancer diagnosis).
Ting et al. 2020 (69)	Malaysia	Prostate cancer, bladder, and renal cancer	2007–2011	429	35.4%	Personal Financial Well-being Scale PFLBS, eight-item Likert scale of 10 points, 1–4 indicate high FT and LWB, 4.1–6.9 indicate average FT, and 7–10 indicate low FT and high financial WL.
Odahowski et al. 2019 (44)	USA	All cancer type	2011	1,419	23.9%	(1) You or anyone in your family had to borrow money or go into debt; (2) You or anyone in your family filed for bankruptcy; (3) You or your family made other financial sacrifices; (4) Unable to cover the cost of medical care visits. those who responded "yes" to at least one of the above questions were defined as experiencing financial hardship.
Ekwueme et al. 2019 (32)	USA	All cancer type	2011–2016	4,753	Material hardship (25.3%) psychological hardship (34.3%)	(1) Material hardship was measured by asking survivors whether they ever had to borrow money, go into debt, file for bankruptcy, or had been unable to cover their share of medical costs. (2) Psychological hardship was considered as being worried about large medical bills.
Bala-Hampton et al. 2017 (26)	USA	Acute myeloid leukemia	2017	26	49.6% Median = 17	COST (COmprehensive Score for financial Toxicity), median 17, less than 17 have distress

(Continued)

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TABLE 1 (Continued)

Author, Year	Country	Cancer type	Year of research	Sample size	Prevalence of FT (%)	Tools used
Aviki et al. 2021 (25)	USA	gynecologic cancer	2021	89	35% Median = 31.9	COST (COmprehensive Score for financial Toxicity) questionnaire, scored <26 experiencing financial toxicity.
Ehlers et al. 2020 (31)	USA	bladder cancer	2020	226	Median = 28.4	COST (COmprehensive Score for financial Toxicity) questionnaire
Durber et al. 2021 (71)	Australia	Thoracic, breast, carcinoma, skin, CNS, Upper GI, gynecological, head & neck, colorectal & urological cancers	2021	257	Median = 26	COST (COmprehensive Score for financial Toxicity) questionnaire
Rosenzweig et al. 2019 (46)	USA	Breast cancer	March–July 2016	145	Median = 23	COST (COmprehensive Score for financial Toxicity) questionnaire
Yap et al. 2020 (68)	Malaysia	All cancer type	2014–2018	461	Median = 22.0	COST (COmprehensive Score for financial Toxicity) questionnaire
Parker et al. 2022 (73)	Australia	Blood cancer	2020–2021	113	Median = 28	COST (COmprehensive Score for financial Toxicity) questionnaire
Albelda et al. 2019 (24)	USA	Blood cancer	June 2014 and January 2015	171	9% answered not at all for Q1, 6% answered extremely difficult for Q2, and 18% answered not enough for Q3	(1) "How satisfied are you with your family's present financial situation?" (1 = completely satisfied and 5 = not satisfied at all); (2) "How difficult is it for you/your family to meet monthly payments on your bills" (1 = not difficult at all and 5 = extremely difficult) (3) "How do your family's finances usually work out at the end of the month?" (1 = some money left over, 2 = just enough money, and 3 = not enough money).
Banegas et al. 2016 (27)	USA	Any type of cancer	4,719	2012	64% reported worrying about having to pay large bills; 34% reported that they or someone in the family had gone into debt because of cancer; 3% of they or their families had filed for bankruptcy; and 40% reported making other financial sacrifices.	(1) Worrying about having to pay large bills related to their cancer, (2) they or someone in the family had gone into debt because of cancer, (3) they or their families had filed for bankruptcy, and (4) making other financial sacrifices
Casilla-Lennon et al. 2018 (29)	USA	Bladder cancer	138		24%	Selecting "agree" or "strongly agree" on the following statement; "You have to pay more for medical care than you can afford" which has 5-point Likert scale options.

(Continued)

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TABLE 1 (Continued)

Author, Year	Country	Cancer type	Year of research	Sample size	Prevalence of FT (%)	Tools used
Gordon et al. 2017 (b) (75)	Australia	Colorectal cancer	187	January 2010 to Septemper 2011	1 to 0.6% answered as poor in 1st domain at 6 and 12 months, financial strain reported by 15 and 7% at 6 and 12 months, difficult to raise money 41 and 33% at 6 and 12 months	FT questionnaire of three domains: perceived prosperity (prosperous, very comfortable, reasonably comfortable, just getting along, or poor or very poor), financial strain (could not pay utilities on time, could not pay mortgage, or rent on time, sold something, went without a meal, unable to heat home, ask for financial help from friends or family, and asked for financial help from an organization) ability to raise money ($2000) (I could easily raise money, unable/difficult to raise money)
Honda et al. 2019 (85)	Japan	All types (Solid Tumours)	2019	156	Median = 21	COST (COmprehensive Score for financial Toxicity) questionnaire
Huntington et al. 2015 (37)	USA	Multiple Myeloma	Between Aug 18, 2014, and Jan 7, 2015	100	Mean = 23.0	COST (COmprehensive Score for financial Toxicity) questionnaire
Kale et al. 2016 (41)	USA	All types	2011 Medical Expenditure Panel Survey (MEPS)	19.6 million	28.7% reported financial burden.	Cancer Self-Administered Questionnaire (CSAQ) Financial burden was present if one of the following problems was reported: borrowed money/declared bankruptcy, worried about paying large medical bills, unable to cover the cost of medical care visits, or other financial sacrifices.
Rogers et al. 2012 (61)	USA	Head and Neck cancer	January and December 2008	447	54% (at least moderate or large financial burden) 34% (had at least 3/17), and 17% (had at least 5/17).	Self-designed questions about the financial burden and benefits are included in the Cost of Head and Neck Cancer Questionnaire. The severity of burden (no burden, little, moderate, large, or not applicable) in relation to 17 different financial issues, and to say which three had the greatest impact because of their cancer. They were asked the level of difficulty (no difficulty, a little, quite a bit, and very much).
Inguva et al. 2022 (39)	USA	All types	2016–2017		53.7	Cancer Self-Administered Questionnaire of the Medical Expenditure Panel Survey, including FT
Sharp and Timmons, 2016 (62)	Ireland	Breast, prostate, and lung cancer	2008	698	48% reported cancer-related financial hardship, and 32% reported strain	Questions were designed to capture objective and subjective measures of financial difficulties. Seven-level Likert-type scales ranging from much more difficult, very concerned, to much less difficult and much less concerned.
Shankaran et al. 2012 (49)	USA	Stage III colon cancer	2008–2010	284	38% of cancer patients had financial hardships, 23% were in debt, with an average debt of $26,860, and 27% had to sell stocks or use savings or retirement funds	Multidimensional survey instrument was used. FT was assessed through four questions: (1) To pay bills related to cancer treatment, have you had to sell house, borrow money, …etc., (2) Any reduction of income, and how much?, (3) f had to borrow money from other friends or family members, (4) Are you currently in debt due to expenses related to your cancer treatment?
Wheeler et al. 2018 (50)	USA	Breast cancer, all stages	2008 to 2013	2,494 women (49% black, 51% white)	58% of black women, and 39% of white women	A modified model from the National Cancer Institute that describes the direct and indirect contributors to adverse financial impact (decrease of income, financial barrier to care, loss of job, loss of insurance, transportation barrier)

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TABLE 1 (Continued)

Author, Year	Country	Cancer type	Year of research	Sample size	Prevalence of FT (%)	Tools used
Whitney et al. 2015 (51)	USA	1,209 cancer survivors	2011	All cancer types and stages except skin melanoma	33.2% indicated financial concerns, with 17.9% reporting debt or bankruptcy, 44.0% of working survivors made work adjustments, and 15.3% of which were long-term	A self-administered questionnaire co-developed by the National Cancer Institute (NCI), American Cancer Society (ACS), Centers for Disease Control and Prevention (CDC), National Institutes of Health (NIH), and LIVESTRONG—to provide national estimates of psychosocial, financial, work-related, and other aspects of cancer burden
Yabroff et al. 2016 (52)	USA	1,202	2011	Not specified	28.4% of patients aged 18–64 13.8% of patients ≥ 65 years old.	Material financial hardship was measured by ever (1) borrowing money or going into debt, (2) filing for bankruptcy, (3) being unable to cover one's share of medical care costs, or (4) making other financial sacrifices because of cancer, its treatment, and lasting effects of treatment. Psychological financial hardship was measured as ever worrying about paying large medical bills

3.5 Cost of cancer management

The cost of cancer management was also reported in the majority of included studies (20, 28, 30, 34–36, 38, 42, 43, 47, 48, 54–57, 60, 62–67, 69, 72–74, 76–84, 86).

3.5.1 Direct medical costs

Data on mean direct medical costs from different perspectives were reported in 39 studies in total (20, 28, 30, 33–36, 38, 40, 42, 43, 47, 48, 53–57, 60, 62–67, 69, 72–74, 76–84, 86). The period during which the expenditures were incurred varied widely in the included studies; some studies calculated the cost among cancer survivors (35, 40, 57), in the past 1 month (80), in the last month of life (28), and 2 years after diagnosis (83). The majority reported the annual cost (30, 33, 47, 48, 54–56, 63, 66, 78). Garaszczuk et al. (2022) found that most of the burden is incurred during the first year after diagnosis, and the most costly cancers are lung, colorectal, and prostate (65). The cost perspective varied widely among studies, with the majority using patient perspectives. However, few studies used the provider perspective or the societal perspective. In addition, some considered the cost of health insurance plans. Detailed cost data are shown in Table 2.

3.5.2 Direct non-medical costs

The direct non-medical cost was included in only seven studies (47, 57, 64, 67, 69, 73, 86). Two studies were conducted on colorectal cancer patients (44, 67): one in prostate cancer patients (64), one among patients of three types of cancer, namely bladder, prostate cancer, and renal cancer (69), one among blood cancer patients (73), and one among patients of any cancer type (86). The cost in the last 3 months for prostate cancer was USD$ 379.38 in Canada (64). For colorectal cancer, one study was done in Malaysia and found the cost to be USD$ 246.8 in the first year after diagnosis, and the other one was conducted in Europe and found the cost to be €510 in the three studied years (2007–2009) (57, 67). Ting et al. (2020) found that the cost per patient is USD$ 24.8 in Malaysia, and Parker et al. found that the cost in Australia is AUD$ 6,700 among blood cancer patients (73). In addition, the cost among cancer patients of any type in Ethiopia was USD$ 1,978 (86), and Sasser et al. (2005) found that the annual cost among breast cancer patients was USD$ 8,236. (Table 2).

3.5.3 Indirect cost

A total of eight studies reported the indirect cost of cancer management (30, 33, 34, 54, 65, 67, 76, 81). Total annual indirect costs in Europe per lung cancer patient were as follows: €696, €2,476, and €1,414 in France, Germany, and the UK, respectively (54). It was USD$ 1,019 compared to USD$ 48 for those with breast cancer with lymphadenoma compared to those without lymphadenoma, respectively, in the USA (30). In addition, the cost was USD$ 452.2 among colorectal cancer patients in Malaysia (67). Moreover, 10 years of indirect costs amounted to CAD$ 2.7 billion among patients of any cancer type in Canada (1097–2007) (65). It accounted for USD$ 19 million in Iran in 2014 among gynecology cancer patients (81). In addition, one study reported the work days missed due to disease rather than the cost of productivity lost (33) (Table 2).

TABLE 2 Direct medical, non-medical, and indirect costs in included studies.

Author, Year	Country	Type of cancer	Year of research	No. of patients	Direct medical cost	Direct non-medical	Indirect cost	Perspective
Chu et al. 2008 (84)	Taiwan	Any cancer site (17)	1990–2001	425,294	Highest lifetime cost per case =2,404,000TWD the highest average annual cost per case = 207,000 TWD	NA	NA	National Health Insurance
Andreas et al. 2018 (54)	Europe	Lung cancer	August 2009 and July 2012	306	Mean cost €19,057 (France), €14,185 (Germany), and €8,377 (UK)	NA	Mean costs per patient were €696 (France), €2,476 (Germany), and €1,414	Societal
Dean et al. 2019 (30)	USA	Breast cancer	2015	40	Annual OOP costs = $2,306 compared to $1,090 for those with and without lymphedema	NA	$1,019 compared to $486 for those with and without lymphedema	Patient
Bao et al. 2018 (28)	USA	Pancreatic cancer-stage IV	2006–2011-last month of life	3,825	Median patient OOP ($1,004.8 vs. $228.5) for patients with vs. without chemotherapy	NA	NA	Patient
de Oliveira et al. 2014 (63)	Canada	Prostate cancer	2014	585	Mean OOP costs were $200/year.	NA	NA	Patient
da Veiga et al. 2021 (77)	Brazil	Skin cancer—all stages	NA	NA	Stage 0:359 and 3,135, stage I: 8022 and 39,345, stage 2: 9365–80,036, Stage III: 12,285–556,983, stage IV: 8070–850,686, in public and private, respectively in Reais (R$)	NA	NA	Healthcare provider
Afkar et al. 2020 (82)	Iran	Breast cancer	2020	76	Total mean hospitalization cost (4343.69 USD) Mean (SD) of patient contributions [281.13 (307.22)]	NA	NA	Societal
Azzani et al. 2016 Azzani et al. 2017 (67, 70)	Malaysia	Colorectal cancer-all stages	2016	138	RM 6544.5 (USD 2045.1) for stage I, RM 7790.1 (USD 2434.4) for stage II, RM 8799.1 (USD 2749.7) for stage III and RM 8638.2 (USD 2699.4) for stage IV	RM790(USD246.8)	USD452.2	Patient
Callander et al. 2019 (72)	Australia	All cancer types	2011–2022	25,553	Direct out-of-pocket 380–1,091 among indigenous and non-indigenous people. Indigenous people spent approximately $269 on healthcare co-payments, and $111 in the 7–12 months post-diagnosis. Non-indigenous people spent $359 in the 7–12 months post-diagnosis.	NA	NA	Patient
Chen et al. 2017 (80)	China	Lung cancer	2017 - past month	227	The mean patient costs were $2518.83. The mean total healthcare cost was $2883.44	NA	NA	Societal

(Continued)

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TABLE 2 (Continued)

Author, Year	Country	Type of cancer	Year of research	No. of patients	Direct medical cost	Direct non-medical	Indirect cost	Perspective
Souza et al. 2011 (78)	Brazil	Skin cancer	2007	42,184:non-melanoma skin cancer cases 2,740: skin melanoma cases	The mean annual cost of NMSC/patient was R$1,172 ± 424 in the public healthcare system and R$1,040 ± 664 in the private system. Melanoma: R$13,062 ± 16,848 and R$26,668 ± 42,750, respectively.	NA	NA	HC provider
De Vrieze et al. 2020 (55)	Europe	Breast cancer	2020 per year	194	Total costs per patient were €2248.9. Within these mean direct costs, €1803.35 (80%) was accounted for statutory health insurance and €445.58 (20%) was out-of-pocket expenses for patients.	NA	NA	Health Insurance and patient
Garaszczuk et al. 2022 (65)	Canada	Any cancer (32)	1997 and 2007	2,000,000	CAD$ 26.2 billion in Canada (2021) from a societal perspective; 30% of costs are borne by patients and families. Patients and families' costs: CAD$ 4.8 billion in 2021.	NA	CAD 2.7 billion	Societal
Hong et al. 2019 (36)	USA	Any cancer	2011 and 2016	655 (2011) 490 (2016)	The mean OOP decreased by $268 (from 384 to 152) after the affordable care act	NA	NA	Patient
Iloabuchi et al. 2021 (38)	USA	Breast, prostate, colorectal cancers, non-Hodgkin's lymphoma	2016	26,822	Mean cost USD 7764	NA	NA	Patient
Lang et al. 2009 (42)	USA	Liver cancer	1999	392	Annual cost: USD$ 454.9 million, Per patient cost: USD$ 32,907	NA	NA	Healthcare provider
Lauzier et al. 2013 (66)	Canada	Breast cancer	2003	800	Median OOP one year after diagnosis is USD$ 1,002	NA	NA	Patient
Mao et al. 2017 (79)	China	Bronchioles and lung, breast, stomach, colon, and rectal cancers	2008	2091	High total expenditure ($1,228) but lowest OOP payment ($170) among the four cities in China (patients with social insurance)	NA	NA	Health insurance and patient
Murphy et al. 2021 (56)	Europe	Breast cancer, genitourinary, GIT, gynecological	Septrember 2018 to March 2019	238	Annual cost: €53,901, per patient: €226.49 Monthly cost: €7,700, per patient: €32.36	NA	NA	Not covered by HC- fundraising, charitable donations, and volunteer and patient

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TABLE 2 (Continued)

Author, Year	Country	Type of cancer	Year of research	No. of patients	Direct medical cost	Direct non-medical	Indirect cost	Perspective
O Céilleachair et al. 2017 (57)	Europe	Colorectal cancer	October 2007–September 2009	497	Average OOP: €1,589 among colorectal cancer survivors	€510	NA	Patient
Parker et al. 2022 (73)	Australia	Blood cancer	April 2020 to February 2021	113	$14,840 among the whole cohort (medication, allied health, and doctor visit)	$6,700 of the whole cohort	NA	Societal
Sargazi et al. 2022 (81)	Iran	Cervical cancer, Ovarian cancer Endometrial Cancer	2014	10,000	$32 million	NA	$19 million	National HC
Sasser et al. 2005 (47)	USA	Breast cancer	1998–2000	555	Average annual direct costs BrCa ($13,925)	$8,236	NA	Employer Medical Claims
Seifeldin, 1999 (48)	USA	Colon cancer	1991–1994	Mean number of admissions: 237,754 per year	Total hospital charge is $4.5 billion per year (4 years period)	NA	NA	National HC
Ting et al. 2020 (69)	Malaysia	Prostate cancer, bladder, and renal cancer	2007–2011	429	USD$ 9181.1	USD24.8	NA	Government subsidy, medical insurance
Vallejo-Torres 2014 (20)	Europe	Skin cancer	2008 and 2020 (estimate)	8,658 Malignant Melanoma and 73,593 NMSC-Year	Range of £106–£112 million in 2008 and estimated to be £180 million in 2020	NA	NA	NHS
Van Agthoven 2001 (60)	Europe	Head and neck cancers	1994–1996	854	£31,829 per patient	NA	NA	HC provider
You et al. 2019 (83)	Korea	Breast cancer	2003–2011	1,087	Mean cost USD$ 12,108 in 2003–2008) after 2 years of mastectomy	NA	NA	National Health Insurance and patient
Kasahun et al. 2020 (86)	Ethiopia	Any type	2018	352	Mean medical cost: $1978 (median: $1394)	Mean cost: $388 (median: $222)	NA	Patient
El-Haouly et al. 2020 (64)	Canada	Prostate cancer	2020	171	The mean total cost incurred in the last 3 months was $517	USD379.38	NA	Patient

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TABLE 2 (Continued)

Author, Year	Country	Type of cancer	Year of research	No. of patients	Direct medical cost	Direct non-medical	Indirect cost	Perspective
Finkelstein et al. 2009 (33)	USA	Any type	2000–2005	1940	OOP during active cancer stage is USD$1,730 and 1,180 in the follow-up stage.	NA	22.3 days	National Health Insurance and patient
Gordon et al. 2017 (74)	Australia	Prostate cancer	April and June 2013	289	OOP median is AUD$ 8,000	NA	NA	Patient
Gordon et al. 2007 (76)	Australia	Breast cancer	2004–2006	287	Mean cost is USD$ 1,937	NA	Mean cost US$6093	Patient
Guerin et al. 2016 (34)	USA	Brain metastasis among lung cancer patients	January 1, 1999 to March 31, 2013	132	Mean cost USD$86,027	NA	Mean cost USD$8,528	Insurance
Guy et al. 2013 (35)	USA	Any cancer site	2008–2010	4,960	Economic burden of cancer is $16,213 per survivor aged 18 to 64 years and $16,441 per survivor aged ≥65 years.	NA	NA	Patient (OOP), Private insurance, Medicare, Medicaid, and other sources
Jagsi et al. 2014 (40)	USA	Breast cancer	2005 to 2007	1,502	Median out-of-pocket expenses were ≤$2,000; 17% of respondents reported spending > USD$5,000	NA	NA	Patient, Health insurance
Sharp and Timmons, 2016 (62)	Ireland	Breast, prostate, and lung cancer	2008	698	Mean direct medical out-of-pocket costs is EURO€1,491	cancer-related costs (mean = €1,180)	NA	Patient

(Continued)

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TABLE 2 (Continued)

Author, Year	Country	Type of cancer	Year of research	No. of patients	Direct medical cost	Direct non-medical	Indirect cost	Perspective
Zheng et al. 2015 (53)	USA	Colorectal, breast, and prostate cancers	2008 to 2012	Colorectal (non-older adult: $n = 169$; older adult: $n = 371$), breast (non-older adult: $n = 777$; older adult: $n = 791$), and prostate (non-older adult: $n = 281$; older adult: $n = 889$) cancer survivors and individuals without a cancer history (non-older adult: $n = 95,640$; older adult: $n = 13,792$)	Annual excess medical expenditures (for the non-older adult population, colorectal: USD$8,647, breast: USD$5,119, and prostate: USD$358; for the older adult population, colorectal: USD$4,913; breast: USD$2,288, and prostate: USD$3,524).	NA	NA	Patient
Massa et al. 2019 (43)	USA	Head and neck compared to other types of cancer	1998–2015	16,771	Median annual medical expenses (USD$8,384 vs. USD$5,978; difference, USD$2,406; 95% CI, USD$795–USD$4,017)	NA	NA	Patient

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4 Discussion

This systematic review and meta-analysis describe the prevalence of subjective and objective FT among cancer patients. The included studies are from different income countries and were published between 1999 and 2022. The majority of studies reported only direct medical costs ($n = 39$). Few studies ($n = 7$) reported direct non-medical costs, which include the cost of transportation, food, and accommodation due to disease, and similar findings were observed earlier (17). Moreover, only eight studies reported the indirect cost among cancer patients, which is defined as the cost of productivity loss of patients and their caregivers as a result of cancer. This was also hardly measured previously (17). Even though cancer care expenses are intuitive indications of the financial effects of cancer care, it was challenging to compare the included research cost findings due to the disparate illness course, type, stage, perspective, and the period during which the expenditures were incurred.

The medical expenditure–income ratio may be more suitable than a particular value for medical costs when evaluating cancer-related FT among patients. However, the definition and methods of measurement were contradictory in the included articles. For example, four studies used CHE to measure the household financial burden of healthcare payments, which is a well-established objective tool (87, 88). It is considered that a patient experiences a catastrophic situation when a household's OOP healthcare expenditure exceeds 40% of the household's capacity to pay (i.e., effective income remaining after basic subsistence needs have been fulfilled) (89). However, one study estimated the CHE using the Wagstaff and Van Doorslaer approach (90); when households with prior-year cancer patients' OOP expenses for care exceeded 10% of their total annual household income, it was deemed catastrophic. As such, we only pooled the prevalence of CHE in four studies and found it equal to (47%) (69, 70, 79, 80), which is less than that found in a study conducted in a low-income country (74.4%) (86). However, this value represents only 11% of the included studies, which reflects that the included studies barely focused on measuring the objective FT.

Regarding the prevalence of subjective financial toxicity, it was found to range between 20.9 and 83.7%. There is a huge variation in measuring the subjective FT among the included studies. This finding confirms the earlier observation that there is a lack of accepted definitions of subjective FT (91). Similar findings have been reported by a previous systematic review, which synthesized methods for measuring FT (8, 17). Recently, a few standardized instruments have been developed and validated in an attempt to quantify the financial toxicity of cancer patients. An example of a COST tool is the de Souza (92) instrument, which was developed in 2014 and validated and used in high- and higher-middle-income countries to measure cancer patients' experiences of financial toxicity. However, it may not apply to lower-middle or low-income countries. The median COST score among included studies [USA ($n = 5$), Australia ($n = 2$), Japan ($n = 1$), and Malaysia ($n = 1$)] ranged between 17 among acute myeloid leukemia and 31.9 among patients with gynecological cancers, in which a low score indicated high financial toxicity (92). Some studies used the median COST score as a cutoff point to define those experiencing FT (25, 26). However, there was a wide variation in the median of included studies, which might require a validation study on COST to standardize the cutoff point to categorize those experiencing FT.

The strength of this study is that it is the first systematic study and meta-analysis to determine the amount of cancer-related financial toxicity and how it has been measured in various income countries. However, this study has several limitations. First, due to the considerable heterogeneity in the outcome measurement utilized in the included studies, our summary of the findings was narrative rather than quantitative (except for CHE). Second, owing to the considerable heterogeneity in the disease period or course during which the costs were incurred, unknowns and inconsistencies in the amount and type of resources included, inflation, and currency rates, we did not synthesize or compare cancer-related expenditures (including medical, non-medical, and indirect costs) across studies.

5 Implication and recommendation

Regular clinical evaluations rarely include FT assessments. According to this review, FT affects cancer patients and their families negatively and is common among cancer patients around the world. As a result, in clinical practice, FT in cancer patients needs to get more attention. The evaluation, acknowledgment, and discussion of financial toxicity are crucial milestones. Nurses can work with doctors to analyze patients' financial burdens and provide information assistance for cancer patients because they have the closest touch with cancer patients and their careers. Therefore, the government, cancer foundations, and other organizations should adopt initiatives such as education and training programs to expand nurses' awareness of FT assessment and patient assistance programs.

More high-quality research is required, especially from low-income nations, on the FT of cancer. A tool to quantify FT in cancer patients has to be developed and validated in further research.

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

MA: Conceptualization, Writing – original draft. WA: Writing – review & editing. DA: Writing – original draft. VK: Writing – original draft. MA: Writing – original draft.

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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.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1266533/full#supplementary-material

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Exploring human resource management in the top five global hospitals: a comparative study

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Background: The pivotal role of Human Resource Management (HRM) in hospital administration has been acknowledged in research, yet the examination of HRM practices in the world's premier hospitals has been scant.

Objective: This study explored how the world's leading hospitals attain operational efficiency by optimizing human resource allocation and melding development strategies into their HRM frameworks. A comparative analysis of the HRM frameworks in the top five global hospitals was undertaken to offer a reference model for other hospitals.

Methods: This research offers a comparative exploration of the HRM frameworks utilized by the top five hospitals globally, underscoring both shared and distinct elements. Using a multi-case study methodology, the research scrutinized each hospital's HRM framework across six modules, drawing literature from publicly accessible sources, including websites, annual reports, and pertinent English-language scholarly literature from platforms such as Google Scholar, PubMed, Medline, and Web of Science.

Results: The analyzed hospitals exhibited inconsistent HRM frameworks, yet all manifested potent organizational cultural attributes and maintained robust employee training and welfare policies. The design of the HR systems was strategically aligned with the hospitals' objectives, and the study established that maintaining a sustainable talent system is pivotal to achieving hospital excellence.

Conclusion: The HRM frameworks of the five analyzed hospitals align with their developmental strategies and exhibit unique organizational cultural attributes. All five hospitals heavily prioritize aligning employee development with overall hospital growth and place a spotlight on fostering a healthy working environment and nurturing employees' sense of achievement. While compensation is a notable performance influencer, it is not rigorously tied to workload in these hospitals, with employees receiving mid-to-upper industry-range compensation. Performance assessment criteria focus on job quality and aligning employee actions with organizational values. Comprehensive welfare and protection are afforded to employees across all five hospitals.

KEYWORDS

world's top hospitals, hospital management, human resource management, organizational culture, value

1 Introduction

Human Resource Management (HRM) navigates through the meticulous planning and rational allocation of human resources, aligning with organizational development strategy by embarking upon a suite of processes like recruiting, training, assignment, evaluation, incentives, and adjustments to maximize personnel value (1, 2). HRM in hospitals emerges as a dynamic key element in the operation and evolution of the facility. Its existence and proficient utilization activate resources effectively, propelling hospitals toward organizational objectives and enhancing healthcare service delivery. Hence, a scientifically sound and efficient HRM becomes indispensable for hospitals to augment their core competitiveness and realize high-quality development (3).

Hospitals, as fundamental entities in the healthcare system, shoulder the vital responsibility of delivering clinical medical services, becoming an essential protective layer for public health (4). The HRM in hospitals, in contrast to other sectors, portrays distinct disparities owing to the specialized service content, clientele, specialized labor divisions, and continuous operation, elevating labor costs and placing specific demands on human resource allocation (5, 6). The heterogeneous and unforeseeable nature of healthcare services also presents notable challenges to internal work process standardization. Thus, maximizing work efficiency through optimal human resource allocation is a vital issue in contemporary hospital management.

The "World's Best Hospitals 2023" has spotlighted top healthcare institutions globally, with the foremost five being the Mayo Clinic (MC), Cleveland Clinic (CC), Massachusetts General Hospital (MGH), The Johns Hopkins Hospital (JHH), and Toronto General - University Health Network (UHN) (7). Exceptional HRM practices are imperative for delivering top-tier healthcare services and achieving remarkable research output. With a pervasive demand for talent in all hospital positions, adopting apt HRM models and strategies to attract, recruit, train, and retain skilled individuals becomes paramount (3, 8–10).

Despite the availability of individual case studies on HRM in some of the world's elite hospitals, there prevails a void in comparative research on hospital HRM models (11–14). This study thoroughly analyzes HRM within the aforementioned medical institutions, identifying pertinent takeaways and extracting insightful models of experience. The objective is to offer a referential framework for other hospitals to enhance their HRM models and strategies."

2 Theoretical foundation

The core components of human resources management encompass planning, pick and placement, professionals, performance, payment and preservation (15). This study focuses on a comparative analysis of hospital human resources management across six modules, which are depicted in Figure 1.

Human resource 'planning' encompasses the meticulous forecasting of human resource demands and supplies essential for realizing the long-term developmental goals of hospitals (15). The 'Pick and Placement' management involves scientifically and judiciously attracting, choosing, and employing individuals in alignment with job prerequisites, ensuring the optimal person is appointed to the correct role at the opportune moment. The concept of 'professionals' entails the employer taking accountability for initial job training and ongoing employee development to acquaint them with their role responsibilities, specific task contents, and skill prerequisites while also consistently nurturing them in accordance with the hospital's career advancement pathways and extensive development plans. 'Performance' management encapsulates the thorough monitoring, analysis, and evaluation of the work processes and outcomes of either departments or individual employees. It strives to refine employee behavior and operational processes to pursue the hospital' s enduring objectives. 'Payment' management incorporates both financial and non-financial rewards and emerges as a pivotal component in achieving incentive and performance management goals. 'Preservation' management predominantly engages with employee performance assessment, employment, and welfare management. Also, it encompasses the crafting of team building within agencies and the generation of organizational culture and ambiance.

3 Methods

In this study, the foremost five hospitals were chosen from 'World's Best Hospitals 2023' (7). The ranking encompasses an extensive list of over 2,300 hospitals across 28 countries and regions, with scoring derived from data involving medical expertise, patient contentment, hospital quality indicators, and patient outcomes.

The primary research methodology employed in this study adhered to a multi-case comparative approach. Engaging in multiple-case research, which involves the comparative analysis of two or more cases under the stewardship of theoretical sampling principles, aims to pinpoint similarities and disparities among the cases under scrutiny and formulate theory (16). When contrasted with single-case research, the multi-case method can more precisely delineate different constructs and their interrelations, pinpointing accurate definitions and appropriate levels of construct abstraction. This approach lays a more robust foundation for theoretical construction and fosters the generation of theories with broader applicability (16, 17). The data for this research was sourced from publicly available information on the five hospitals, including official websites, annual reports, and pertinent English academic literature on platforms like Google Scholar, PubMed, Medline, and Web of Science (WOS). It is pivotal to note that the research materials were derived from public domains and did not engage in any biological research involving humans or animals.

4 Results

The five hospitals are all non-profit organizations with a history of over 100 years. MC, CC, MGH, and JHH are in the United States, while UHN is in Canada. Four hospitals are private institutions except for UHN. The basic information of the five hospitals can be found in Table 1.

4.1 Planning

As shown in Table 2, each hospital has unique characteristics in human resources planning. At MC, employee cost analysis is performed and information systems are used for business prediction to clarify the hospital development scale and corresponding personnel needs. Remote full-time positions are created based on job content to overcome the spatial limitations of the workplace, improve work efficiency and output quality, and incorporate human resources planning indicators into hospital development strategies. At CC, human resources planning is independently borne by the medical group, with dedicated management departments responsible for personnel recruitment, cost control, and hospital profit and loss. MGH has "The Massachusetts General Physicians Organization" serving as an independent organization responsible for the appointment and removal of medical personnel and the development and modification of related rules and regulations. UHN formulates human resources strategies and job responsibilities based on the hospital's development vision and goals, with a highly-developed employee welfare system that encourages employee participation in hospital management practices. At JHH, a data-driven approach is adopted for human resources planning and analysis, using personnel analysis, labor analysis, or talent analysis as the basis for personnel management to improve decision-making.

FIGURE 1
Six modules of human resources management.

4.2 Pick and placement

Table 3 presents the recruitment and staffing features of each hospital. MC follows the "values first" principle and emphasizes the alignment of values by selecting candidates who embrace Mayo's value "The needs of the patient come first" through at least two rounds of interviews. Candidates are selected through at least two rounds of interviews and are regularized after an initial probationary period and an in-depth probationary period of about 3 years. A variety of corporate culture training programs are also in place to emphasize diversity in talent selection and to increase the percentage of minorities among employees.

To avoid conflicts between management decisions and the best medical decisions, management positions at CC are filled by technical professionals with medical backgrounds. When recruiting managers, candidates are screened by management and are required to choose between management and clinical career paths, and are recognized by

TABLE 1 Basic information of the five hospitals.

Hospital	MC	CC	MGH	UHN	JHH
Ranking 2023	1	2	3	4	5
Headquarters	Rochester, Minnesota, United States	Cleveland, Ohio, United States	Boston, Massachusetts, United States	Toronto, Ontario, Canada	Baltimore, Maryland, United States
Since	1883	1921	1811	1829	1889
Care System	Private	Private	Private	Public	Private
Beds	1,243	-	927	-	-
Number of Employees	73,600	-	23,173	-	10,400
Beds-to-Employee Ratio	0.017	-	0.04	-	-
Annual Outpatient Cases	Over 1.4 million	-	1,440,548	-	-
Campuses & Branches	3 & 2	5 & 4	1 & 0	3 & 2	1 & 0
University/Affiliated	Mayo clinic school of medicine	Cleveland clinic lerner college of medicine	Harvard university	University of Toronto	Johns Hopkins university

TABLE 2 Characteristics of human resources planning in five hospitals.

Hospital	Characteristics
MC	Human resources planning indicators are datamined and integrated into the overall hospital strategy
CC	The medical group is independently responsible for the planning
MGH	Managed by the Massachusetts General Hospital Physicians Organization as an independent institution
UHN	Developed in line with the hospital's vision and goals
JHH	Adoption of a data-driven approach to human resources planning.

TABLE 3 Characteristics of human resources pick and placement in five hospitals.

Hospital	Characteristics
MC	1.Equal emphasis on professional skills and value consistency; 2. Focus on the diversity of backgrounds of talents
CC	Management positions are filled by skilled professionals with medical backgrounds to minimize management decision-making errors
MGH	The Personnel Specialist assists section managers in the implementation of specific human resources management tasks
UHN	Use of the Internet, partnerships with colleges and universities, and student recruitment programs
JHH	Recruitment and onboarding of employees managed by the HR team

management before entering management. From the management to the medical staff, all of them have specialized medical knowledge. With a clear division of labor in the team, doctors and nursing staff can be exempted from involvement in matters other than therapeutic care, reducing the risk of medical corruption (18).

The recruitment and staffing of MGH is the responsibility of the leaders within the Massachusetts General Hospital Physicians Organization (MGHPO), in conjunction with the hospital president. Departments and physicians are supported by secretaries for day-to-day management to increase efficiency (19). Each department has an administrative director with a professional management background in addition to the department chair. The administrative director is responsible for the development strategy, finance, personnel, and operation of the department. In terms of human resource management positions, the personnel commissioner model and the departmental responsibility system are adopted to achieve a high degree of compatibility between human resource management and departmental management.

UHN utilizes various means such as Internet recruitment, university collaborations, student recruitment programs, and other strategies to broaden the applicant pool and widely attract top talent (20). Full-time recruiters are established to address job vacancies, collaborating with educational experts to ensure a talent pipeline and bridging the gap between hospital talent needs and university training/recruitment efforts (21). Its student recruitment program, which includes co-op, summer, internship, and post-graduate programs, involves nine colleges and universities that fulfill the hospital's need to hire more students to address short-term, high workloads and projects in addition to traditional full-time recruiters. JHH follows a traditional, centralized HRM model, with the Career Services team in the Human Resources Department handling all new hires in a centralized manner.

JHH uses a traditional centralized HRM model, with the Career Services team in the Human Resources Department handling the recruitment and on-boarding of its new employees.

TABLE 4 Characteristics of human resources professionals in five hospitals.

Hospital	Characteristics
MC	1.Provide training opportunities for all employees; 2. Use evaluation indicators to test the results of employee training.
CC	1.Diversified course options, combining online and offline, medical and non-medical educational activities; 2. Training courses in cooperation with external institutions
MGH	1.Each staff member is required to receive training and pass an assessment; 2. Set up a career development center to provide staff with guidance and assistance in all areas according to their needs; 3.Set up a dedicated department to ensure that staff personal growth goals and departmental development goals are harmonized; 4.Arrange for managers to receive leadership training
UHN	1.Establishment of mandatory online learning courses; 2. Structured career paths
JHH	1.Job-related training is conducted by the Office of Human Resource Management Strategic Development; 2. REACH helps employees gain the skills and knowledge needed to fill vacant positions

4.3 Professionals

The characteristics of training and HRD in each hospital are summarized in Table 4. MC provides various training programs and learning opportunities for all employees, emphasizing the training of technical staff with humanistic content such as leadership and career goal planning. All new employees undergo training on the values of "The needs of the patient come first." Each healthcare worker is given the opportunity to become a manager in the corresponding department or division, and evaluation indicators are introduced to monitor the results of staff training.

CC places a strong emphasis on personnel training, offering more than 50 courses each year internally, which have been attended by a cumulative total of over 7,000 professionals and technicians. The education and training of medical professionals and technicians is carried out through collaborations with several universities and organizations both domestically and internationally, as well as other medical institutions. For example, continuing medical and non-continuing medical education activities are conducted by the Center for Continuing Education in cooperation with the Institute for Quality and Patient Safety; continuing medical education is conducted through online learning and on-site regular educational activities. Special communication experience courses are offered, leadership

training is provided for medical staff who do not have a management background, and a professional management team is set up to assist in administrative work, ensuring that professionals and technicians can remain focused on clinical practice.

MGH regards its staff as the hospital's greatest asset, has a comprehensive training system, and adheres to the principle that no one can be employed without completing the training program. Its Career Development Center includes the Clinical Career Development Office, the Research Staff Development Office and the Women's Development Office. It provides daily training and is responsible for the annual Career Development Conference. Through the Annual Career Conference, employees' personal expectations are aligned with their growth goals and departmental development goals, enabling them to better serve the organization's common interests while achieving good development in their personal careers. Each department discusses with employees their personal growth goals and departmental development goals to find common ground and make work plans and career plans for the following year. Guidance and assistance are provided to employees according to their needs, and a variety of scholarships and grants are available to help alleviate the financial burden of learning new knowledge and skills, such as the Tuition Assistance Program. Managers need to receive leadership training. A "Management Trainee" program is offered to provide 2 years of training to promising talent. Interns are assigned to different departments for internships and can also join the hospital's senior operations team to learn and practice.

UHN has separate mandatory e-learning courses for different categories of employees, and designs structured career paths that provide clear opportunities for advancement for individuals with the right skills and experience. A professional development budget is established for course fees, book purchases, and conference planning in the areas of project management and application support. Employees who receive educational development grants are required to submit appropriate training reports for review by all department members. Team-building workshops are held to encourage employees to share their expertise with colleagues (22).

JHH's Human Resources Department develops training programs based on the needs of the organization and integrates on-the-job training with the organization's performance goals. The Office of Strategic Workforce Development (SWD) provides advice and assistance to employees in identifying next steps and developing plans to achieve their goals. SWD through the Hopkins Resources and Education for Career Development (REACH) program, provides the JHH system's SWD provides in-person and online career guidance to individuals and groups through the Hopkins Career Development Resources and Education (REACH) program, which identifies appropriate openings for current employees and provides career development counseling to current employees and community adults and youth, and participates in this effort in partnership with Johns Hopkins University.

4.4 Performance

The characteristics of each hospital's performance management can be seen in Table 5. The value concept of MC runs through the whole process of its human resource management. The purpose of performance appraisal is to help employees improve their competence and is not linked to salary or bonus. The performance appraisal includes three areas: medical care, medical education and medical research (the triple shield). The employee's annual performance review consists of a self-assessment and a supervisor's evaluation. The appraisal includes the core values of each position, required professional competencies, achievement of annual goals, and an overall assessment of accomplishments. The results of the performance review are used as a guide for ongoing employee training rather than as a basis for termination (23). Integration of HR assessment and financial performance evaluation with organizational development strategies using HR balanced scorecard, human capital return on investment metrics with non-profit strategy maps. Diversified incentive models such as spiritual rewards (e.g., Karis Quarterly Awards) and academic evaluation systems were used to comprehensively assess healthcare workers' contributions.

TABLE 5 Characteristics of human resources performance in five hospitals.

Hospital	Characteristics
MC	1. Performance appraisal has nothing to do with workload, and the results of performance appraisal serve as a basis for hospitals to develop skills for their employees; 2. Enhance employee performance through diverse incentive models
CC	1.A comprehensive performance evaluation system; 2. Create special awards to recognize exemplary service and dedication to excellence
MGH	Multiple awards to recognize outstanding employees
UHN	Establishment of special awards to recognize performance in daily work in line with organizational values
JHH	1. use an electronic performance appraisal system; 2.Set up employee appreciation and recognition programs

The most important feature of CC's human resource management model is its high-frequency and flexible performance appraisal approach. Measures such as the development of implementation processes, the creation of evaluation charts, and performance appraisals ensure the continuous progress of the organization, and CC healthcare workers can plan their work and allocate their time according to their personal wishes to achieve optimal delivery of their work through a high-frequency evaluation cycle once a year. There is Maria and Sam Miller Professional Excellence Award that recognizes exemplary service and dedication to excellence.

MGH has adopted a cultural assessment model for year-end appraisals, with high performance ratios to motivate employees and integrate their values into the organization's operations, such as the "icare in Action" program that recognizes individuals and teams who have been singled out in patient thank-you letters. The annual Popular Patient Experience Award recognizes extraordinary dedication, and recipients are honored at an annual ceremony hosted by senior executives.

UHN takes a similar approach to performance assessment, such as the "Living Our Values Awards," which recognizes outstanding employees by the results of living the organization's values (safety, caring, teamwork, integrity, and stewardship) in their daily work.

JHH uses an online electronic performance management system in conjunction with Johns Hopkins, and human resources departments provide department heads with the tools and resources they need to

TABLE 6 Characteristics of human resources payment in five hospitals.

Hospital	Characteristics
MC	Adoption of a fixed annual salary system to motivate employees with diversified non- material incentives and welfare policies
CC	Healthcare workers' earnings are not related to bonuses or workload
MGH	Higher salary expenditure as a percentage of total expenditure
UHN	Establishment of a dynamic pay monitoring and adjustment strategy
JHH	Employee salaries are higher than the average income level of peers

conduct performance reviews, assisting with departmental payroll and providing them with the tools and resources they need to conduct performance reviews. Tools and resources to assist in departmental salary planning decisions. Employee appreciation programs are also set up to strengthen the positive guiding role of organizational culture.

4.5 Payment

The compensation management of each hospital is characterized as follows in Table 6. MC has a pre-determined target annual salary based on job position, with equal pay for equal work, and it does not have a pure salary system based on performance incentives. Compensation is determined based on a combination of an individual's contribution to the organization and their competence rather than job position. Regular reviews and internal satisfaction surveys are conducted by the Remuneration Officer. Remuneration levels are higher than the industry average (at the 60th percentile). MC has adopted a fixed annual salary system that decouples performance from salary income and uses remuneration, as well as diversified non-material incentives and benefits policies, to provide employees with motivation (24).

CC healthcare workers' income is not linked to the volume of treatment, and its salary and benefit expenses account for 56% of the center's operating income, with this high proportion ensuring that healthcare workers' income remains in the upper middle of the industry within its strict payroll system (25). MGH operates a relatively similar compensation mechanism to CC, with employee salary expenses accounting for approximately 42% of total expenses (26).

UHN utilizes external benchmark for pay adjustments, using benchmark jobs and extracting market salary data at the 50th and 75th percentiles. In-depth research was also conducted on hospitals, leading to the benchmark of pay levels in five hospitals with a dynamic adjustment mechanism in place to ensure that their pay levels were appropriate. Staged salary reviews and adjustments were conducted every 3 months, and market analysis reviews and adjustments were conducted every 6 months to ensure fairness by modifying pay policies for overtime, return to work at any time, and shift work (27). A relatively independent and centralized personnel management model has been adopted at JHH. Employee payroll is accounted for and paid by the Payroll Shared Services Division of the Human Resources Information Processing Center (HRIPC).

TABLE 7 Characteristics of human resources preservation in five hospitals.

Hospital	Characteristics
MC	Ensure high levels of employee satisfaction to improve talent retention rates. Focus on high turnover employee groups, identify the key causes, and take corrective actions. Provide employees with a comprehensive benefits package.
CC	Conduct of annual staff-wide evaluations and annual reassessments and appointments
MGH	1.Free and Confidential Employee Assistance Program to help employees achieve work-life balance; 2.Generous Employee Benefits Package
UHN	Performing Motivational Analysis on Departing Employees for Continuous Improvement
JHH	1. prioritize identifying and retaining talent while fostering a strong sense of organizational identity among employees; 2. implement employee assistance initiatives

4.6 Preservation

The characteristics of each hospital's labor relations and benefits management are summarized in Table 7. MC assesses employee satisfaction through employee surveys and third-party evaluations with the aim of reducing turnover rates through high levels of employee satisfaction. Special attention is paid to highly mobile employee groups, such as registered nurses, and in-depth studies are conducted through focus group interviews and other means to identify key causes and take remedial action. Employees are provided with a comprehensive benefits package including medical, dental, tuition reimbursement, paid leave, and defined benefit pension plans, etc. CC signs one-year term contracts with all employees, including top management, conducts annual reassessments and appointments, conducts annual professional reviews, and adopts an accountable model of contractual appraisal.

MGH operates a free and confidential "Employee Assistance Program" to help employees achieve work-life balance; it is not only a counseling office but also an online community called "myStrength" where employees can find solutions to their problems and interact with other members of the community; and a generous employee benefits package that includes healthcare, retirement benefits, employee wellness program activities, childcare centers, consumer discounts, pet insurance, and transportation subsidies.

UHN conducts surveys for departing employees to explore the reasons for their departure and analyze their motivations, including salary, workload, career opportunities, and professional development (28). JHH has an Applause program for employee recognition, which consists of several components, including service anniversary recognition, peer recognition, manager recognition, and birthday recognition. Similar to MGH, JHH has an "Employee Assistance Program" (JHEAP) to help employees through difficult stages.

5 Discussion

As the world's top hospitals, these five hospitals undoubtedly have some commonalities and differences in human resource management

concepts and practices. Examining the similarities and differences while analyzing the potential causes or effects requires consideration of the following six modules of human resource management.

5.1 Planning

All five hospitals have integrated the overall strategic direction into their human resources planning. Both the MC in the United States and UHN in Canada have conducted HR planning analyzes based on data indicator tools. Compared with the MC's analysis system, which is more refined and covers indicators of different dimensions such as employee cost analysis and return on human capital investment indicators, these tools provide sufficient rationales for human resources decision-making and enhance the scientific nature of human resources planning. MC, CC, and MGH will be responsible for the human resources planning functions from the medical institutions, medical groups, and administrative teams that are responsible for the relevant work. This approach enables medical professionals and technicians to focus on the diagnosis and treatment of patient services, while professional managers carry out human resources planning and daily management. Human resources planning needs to be integrated with the organization's development strategy to maintain consistency in management implementation. The concept of strategy first is adhered to in manpower planning, and the medium- and long-term development plan of the institution is used as a starting point to determine the appropriate organizational structure and team model for human resources planning.

5.2 Pick and placement

In terms of recruitment and staffing, the MC ensures that employees who share the organization's values are selected and retained by means of a values-based recruitment philosophy and a long recruitment, training, and probationary period designed to foster the organization's long-term growth and vision. MC departments are managed through a "partner leadership" system, in which physician specialists and professional managers work together to make management decisions. The effective functioning of this model depends on the degree of coordination between the physicians and managers, who need to focus on patient care, and the managers who need to be accountable to shareholders and ensure that the organization operates efficiently and appropriately in terms of revenue. On the other hand, UHN achieves an adequate supply of quality human resources through programs such as the Student Recruitment Program, which is closely related to its status as a teaching hospital of the University of Toronto. The departments and staffing at the five hospitals are quite different. In general, hospitals will tailor their recruitment strategies and employee retention measures according to their specific organizational culture, strategic goals, and resource allocation.

MHG's "dual director" system is similar to the MC's "dual-track" mechanism, with the department director responsible for medical, academic, and other professional areas, and the administrative director responsible for departmental development strategy, finance, personnel, operations, and other work. This approach helps to avoid the department director being trapped in complex administrative affairs and unable to focus on business work, while also assisting in realizing the specialization and refinement of management. CC uses personnel with medical backgrounds to ensure that decisions do not deviate from specialized medical knowledge. At the same time, through clear division of labor in the team, medical staff can focus on clinical diagnosis and treatment and nursing care, which is similar to Mayo's practice. JHH has a relatively centralized human resources management staffing structure, with a vice president in charge of different areas of the organization, including a director of pensions, a human resources consultant, a director of human resources development and training, a director of recruiting, and a director of compensation and benefits. MGH has a management specialist in charge of all types of human resources management, enhancing the ease of interface with human resources services for departments and employees.

The five institutions in this study do not have the same organizational structure and team model, but they have all achieved outstanding development results, demonstrating that both traditional departmental settings and innovative hospital structures can achieve quality development, but must be accompanied by appropriate management models. Human resource planning based on the organization's strategy can maintain consistency in management at the same stage and avoid rotation in key positions.

5.3 Professionals

In terms of staff training and human resources development, all five hospitals attach great importance to the continuing education and diversified training of their staff. The content of the training is unique, but all of them involve the development of comprehensive qualities in various aspects rather than being limited to clinical skills. Many hospitals also emphasize leadership training programs: MC focuses on improving the leadership, goal-planning, and other personal qualities of professional and technical staff; CC offers various leadership programs for clinical staff; MGH focuses on training for managers; JHH implements leadership development through courses, tasks, and other multidimensional means. Leadership is a quality that should be possessed by all types of workers and contributes to superior performance and operational outcomes in hospital organizations.

In addition, the curricula of the five hospitals are unique: Mayo Clinic emphasizes training on values, Cleveland Clinic offers communication experience courses and has built a comprehensive online training system, MGH customizes training programs for individual needs, UHN encourages peer-experience-sharing-oriented learning through a series of seminars, and JHH customizes its training programs from the perspective of talent identification and development plan development. It is worth noting that each hospital places considerable emphasis on the career planning and development of individual employees. Mayo Clinic uses an evaluation and monitoring system to track employee development, MGH helps employees adjust their plans through an annual career development meeting system, UHN has a separate career development budget to provide career path options for employees, and JHH helps employees formulate their career development plans through a comprehensive assessment. It can be seen that the development of an organization cannot be separated from the development of each individual; the two

are complementary, and an excellent organization will do its best to help rather than limit the development of individual members.

Prioritize the development of individuals with diverse expertise, fostering administrative teams that function effectively in both managerial and clinical capacities, while also elevating the professional competence of the leadership knowledge of clinical medical professionals. The allocation of human resources in hospital management positions directly affects the overall level and efficiency of hospital management. Because hospital management is in the interdisciplinary field of medicine and management, the output of relevant professionals is relatively small, many hospitals are still lacking in specialized management personnel, mostly doctors and nursing staff directly, resulting in the de-professionalization of hospital management, and this kind of sloppy management brings about the disorder of hospital governance. Therefore, the optimal allocation of human resources for hospital management positions can be achieved by introducing and training professional hospital managers and improving the management literacy of clinical workers.

5.4 Performance

In terms of performance management, MC's performance management model is different from the other four hospitals and has its own system. Firstly, it divides performance appraisal into five levels from macro to micro, i.e., strategic performance - corporate performance - the triple shield performance (medical care, medical education and medical research) - the performance of various medical services - employee performance, thus realizing the organic link between individual employee performance and the overall organizational performance. Secondly, MC combines individual performance appraisal with performance appraisal of overall HR work through various quantitative indicators and a values-based multidimensional appraisal system, the results of which are used for feedback to individual employees and hospital management as follow-up guidance.

It is worth noting that all five hospitals have established non-material rewards as an important part of employee performance evaluation. For example, MC has the Karis Quarterly Award, CC has the Maria and Sam Miller Professional Excellence Award, MGH has the Annual Patient Experience Award, UHN has the Living Our Values Award, and JHH has the Employee Appreciation and Recognition Program. Different types of non-material rewards are established to recognize the outstanding contributions of employees from different perspectives and to give excellent employees a sense of spiritual satisfaction so that they can execute their quality beliefs more firmly. At the same time, the establishment of awards targeted at teamwork and evaluation by others is also conducive to enhancing the cohesion among employees.

With cultural guidance as the core complemented by the use of scientific management tools, emphasis is placed on the shaping of corporate culture. The five hospitals studied all have distinctive corporate cultures and values, and their cultural infiltration is reflected in all aspects of human resource management. There is even a lack of performance evaluation systems that use cultural identity as an assessment indicator. The advantage of cultural guidance is to ensure consistency of internal values, ease in implementing management decisions, and further ensure the quality of medical services through consistency of values and teamwork harmony. However, the formation of corporate culture takes a long time and places high demands on core managers, making it difficult to form in a short period of time and easy to assess. Therefore, it is necessary to introduce the use of scientific management tools. Management tools serve as an objective evaluation mechanism combined with cultural guidance. Before the formation of a stable and positive corporate culture, the system serves as an objective assessment mechanism; in a stable and positive corporate atmosphere, it can also be used as a management supplement, ensuring fairness and objectivity of the assessment while reducing the subjectivity of the assessment caused by changes in management personnel.

5.5 Payment

In terms of compensation, the fixed compensation of MC and CC lacks the incentive effect of "more work, more pay," but it has not caused slackness in work and lower quality of work. This is because the attractiveness of the clinic's own work platform and the competitive level of salary offered enable employees to obtain reasonable returns. On the other hand, the two clinics also use other means to make their employees unwilling or afraid to relax their work requirements. MC adopts more mental incentives and a comprehensive welfare program, while CC uses contractual appraisals to make employees take their work seriously. The pay adjustment system of UHN follows a cyclical approach to ensure that real-time dynamics allow employees to receive pay in line with the market level, thus ensuring the scientific nature of pay setting, which neither over-motivates nor avoids the lack of motivation caused by under-motivation.

To emphasize humanistic management and non-economic rewards, upholding the concept of humanistic care while focusing the needs of employees. The five healthcare organizations in this study view employee welfare as an important part of human resource management. Not only is career development an important element, but there are also corresponding welfare measures and protection mechanisms for their personal and family members' education, health, and economic status. As a talent-intensive industry, healthcare employees are highly educated and value personal development more than financial compensation. The five organizations have invested significant human and material resources in building a high-quality employee welfare system. For example, the Employee Assistance Program of MGH was designed to help employees balance work and life, creating a good environment for employees to be able to devote themselves to their work. Employees can seek help from the program office for any problems they may have. There are strong professional barriers in the medical services industry, with both the organization and patients seeking the purity of medical services. High-quality employee services are provided to employees to solve their concerns, while ensuring the simplicity of medical services and avoiding the impact on patient safety and treatment effects due to additional performance issues.

5.6 Preservation

Employee satisfaction and turnover are indicators that several hospitals focus on in different ways. Mayo Clinic comprehensively

understands employee satisfaction through surveys, staff meetings, third-party observations and other means, and analyzes the motivation for leaving through focus group interviews and other research methods. Toronto General Hospital conducts a similar analysis of reasons for leaving and uses it to develop talent retention strategies. Loss of professional talents is a bottleneck problem facing all regions and types of hospitals, and due to the long cycle and high cost of training medical professionals, the loss of talents to hospitals is extremely serious, so talent retention is an important issue of concern to the world's leading hospitals.

These hospitals place great emphasis on employee welfare and have designed detailed welfare programs that consider various aspects of employees' lives and work, enabling them to work with peace of mind. Although these welfare programs may seem to increase the cost of hospitals, they are significant in terms of improving employee efficiency and sense of belonging. Soft benefits not only compensate for limitations in salary but also reflect the "people-oriented" value concept, making employees feel the hospital organization's care and support for individuals, and thus more motivated to perform their work. In terms of labor relations management, CC adopts a yearly contract evaluation model and renewal of employment on an annual basis, which avoids to some extent the loss of a sense of crisis and stability brought about by long-term or open-ended contracts, and necessitates all types of employees to maintain a rigorous and conscientious attitude toward their work.

5.7 Strengths and limitations

This study provides an in-depth analysis of HRM practices within the specified medical institutions, discerning key insights and deriving valuable models from their experiences. It proposes a framework as a reference for other hospitals looking to bolster their HRM models and strategies. However, it is essential to acknowledge certain limitations within this study. One such limitation pertains to the representativeness and generalizability of the sample cases, which necessitates further enhancement. The applicability of these cases to HRM practices in hospitals across diverse nations and systems also requires additional exploration.

Furthermore, the literature review is constrained by information sources, making it challenging to accurately and comprehensively reconstruct human resource management practices in various hospitals compared to field research and alternative research methodologies. The content differences in the public data of these hospitals make it challenging to acquire more comprehensive details about their human resource management systems and the actual implementation of these systems. Therefore, there could be potential discrepancies resulting from variations in the disclosure of information across healthcare organizations. More comprehensive information will be obtained through field research or in-depth interviews for comparative studies in the future. It is the hope that future research endeavors will see more researchers and teams contributing to empirical research outcomes in the global field of hospital human resource management. This would further refine and augment resources for hospitals worldwide, aiding in improving their HRM endeavors and realizing high-quality development.

However, the overall ranking of the journals is based on international recommendations from peers in different countries and incorporates the hospital scores. Therefore, the top five hospitals in the ranking are comparable.

6 Conclusion

The study provides an in-depth analysis of Human Resource Management (HRM) practices within five top-ranking global hospitals, unveiling both universal and unique strategies to pave the way for innovative HRM in other hospitals. The insights underscore the critical alignment of human resources planning with an organization's developmental strategy, ensuring management consistency and strategic personnel placement. Success has been witnessed through both traditional and innovative organizational structures when paired with apt management models. A distinctive corporate culture and value system have been pivotal, although their formation demands considerable time and managerial input, highlighting the need for scientific management tools for objective evaluation. A significant focus is placed on nurturing multidisciplinary talents and creating cohesive administrative teams, addressing the current deficit of specialized management personnel in many hospitals. The analysis aims to provide a blueprint for improving HRM strategies in healthcare institutions worldwide, considering the divergences and similarities in HRM practices across eminent hospitals. Future investigations should explore how specific information technologies have been used to promote the effectiveness and efficiency of human resource management in large hospitals.

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 authors.

Author contributions

XW: Writing – original draft, Data curation, Methodology. RW: Conceptualization, Data curation, Writing – original draft. XQ: Writing – review & editing, Data curation, Investigation. Y-NH: Data curation, Writing – review & editing. H-CC: Writing – review & editing, Conceptualization, Methodology, Project administration, Supervision. B-LW: Conceptualization, Methodology, Project administration, Supervision, Writing – review & editing, Funding acquisition.

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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.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2023.1307823/full#supplementary-material

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Incentive mechanism for sharing and using EHR in medical consortiums based on performance evaluation

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Background: The construction of medical consortiums not only promotes active cooperation among hospitals, but also further intensifies active competition among them. The shared use of electronic health records (EHR) breaks the original pattern of benefit distribution among hospitals.

Objective: The purpose of this paper is to establish an incentive mechanism for the shared use EHR, and to reveal the incentive effect and mechanism of key factors, and to put forward management suggestions for solving the real conflicts.

Methods: We constructed a basic incentive model and an incentive model that introduces performance evaluation as a supervisory signal, based on analyzing the hospital cost function, the hospital benefit function, and the incentive contract function. Finally, the incentive effects of key factors before and after the introduction of performance evaluation were verified and compared using MATLAB simulation method.

Results: The profit level and incentive coefficient of hospitals sharing EHR are independent of the amount of one-time government subsidies. Regardless of whether a performance evaluation supervisory signal is introduced or not, the incentive coefficients are increasing functions with respect to ρ, τ, but decreasing functions with respect to β, δ, γ. After the inclusion of supervisory signal of performance evaluation in the model, the ability of hospitals to use EHR has a higher impact effectiveness on improving both incentive effects and benefit levels. The impact of the value-added coefficient on the level of earnings is consistently greater than it would have been without the inclusion of the performance evaluation supervisory signal.

Conclusions: Enhancing the capacity of hospitals to use EHR and tapping and expanding the value-added space of EHR are 2 key paths to promote sustainable shared use of EHR. Substantive performance evaluation plays an important role in stabilizing incentive effects.

KEYWORDS

electronic health records (EHR), medical consortiums, incentive mechanism, performance appraisal (PA), health information

Introduction

A medical consortium is usually composed of public hospitals at different levels within a specific region, and is a consortium that provides primary care, two-way referral, emergency and slow treatment, and up and down linkage of diagnostic and treatment services. While the construction of the medical consortiums promotes active cooperation among hospitals, it also further intensifies proactive competition among hospitals. However, the shared use of EHR resources will break the original pattern of benefit distribution among hospitals. Patients who originally chose to seek medical treatment at this hospital may choose to seek medical treatment at another hospital. Which will further exacerbate the fierce competition among hospitals, thus hindering the realization of the goal of building medical consortiums (1). The integration of electronic health records (EHR) in medical consortiums is divided into two levels: the interconnection of infrastructures such as software and hardware, and the shared use of EHR resources. Interconnection is not the end, shared use is the only way to create value. To reduce the "free riding" phenomenon in the integration of EHR and promote the active sharing and use of EHR resources, it is necessary to establish an effective incentive mechanism that coordinates the positive actions of all parties.

Incentive mechanisms research is the core proposition for solving problems such as alliance cooperation, multi-agent collaboration and collaborative innovation. Incentive mechanism research is also an effective method to study the path of activating the motivation of participating subjects, which has been widely applied in the fields of supply chain management (2), online marketing (3), venture capital (4), benefit distribution (5), information sharing (6, 7), data quality (8), and innovation dynamics (9, 10). In the field of healthcare, incentive issues have always been a major focus and difficulty in practice. However, incentive mechanism research has not been paid much attention to until the beginning of this century in China, with a view to fully stimulating the enthusiasm and innovative vitality by clarifying incentive factors and revealing the mechanism of driving forces. Li and Zheng (11) studied the incentive and constraint mechanism in the coordination of the conflict between individual and collective interests in the construction of county medical communities. Xiong et al. (12) took Sanming Medical Community as the case to reveal the economic incentive mechanism in the supply of medical services. Fu (13) focuses on the incentive mechanism of medical insurance payment reform for physicians.

EHR sharing within the consortium breaks through the limitations of information sharing within individual hospitals. Its core essence is the sharing and interoperability of diagnostic and treatment information between hospitals. Cross-organizational healthcare information sharing has been recognized as a typical scientific problem for future research (14). However, most current researches mainly start from the technical perspective, exploring the system architecture and key technology solutions for information sharing between hospitals (15–17). Some scholars have also begun to pay attention to sharing risks (18), sharing willingness (19), privacy protection (20), sharing models (21), service models (22) and influencing factors (23). In recent years, research on the incentive mechanism for sharing medical and health information, with electronic health records as the core, has gradually received attention from the academic community. For example, Li et al. (24) used a game approach to analyze the incentive mechanism for sharing hospital diagnosis and treatment information. A small number of scholars, such as Jiang et al. (25), have also paid attention to and explored this topic. However, academic research on incentive mechanisms in the shared use of EHRs is far from adequate, with a small number of relevant studies and a depth of research that needs to be further improved.

In view of the above, this paper constructed a basic incentive model and an incentive model that introduces performance evaluation, respectively, based on the principal-agent theory. The model results were solved and the incentive effects ware comparatively analyzed using numerical simulation. The significance of this research is mainly reflected in three aspects: first, it reveals the incentive effect and mechanism of key factors in promoting the shared use of EHR; second, it further proves the important role of performance evaluation in the design of contractual incentive mechanism; and third, it provides decision-making reference for the practice of integrating EHR in medical consortiums.

Methods

Basic incentive model construction

Cost function of hospitals

Hospitals may need to add equipment and modify interfaces to achieve EHR sharing. In order to make EHRs directly available to other hospitals after sharing, it is necessary to integrate data and optimize the format. This inevitably requires investment in manpower and resources, which entails a certain cost. We refer to this cost as the shared cost. The amount of shared costs invested is closely related to the hospital's effort, which can be characterized by the effort level. The effort level can be expressed as $\frac{1}{2}\beta M^2$ (26), where β represents the cost coefficient of hospital investment, and M represents the hospital's earnings level (benefit level). There are also certain risks for hospitals in the EHR integration. These potential risks may include reduced revenues due to interoperability of test results, and patients loss due to convenient access to medical care. Such potential risk cost can be expressed as $\frac{1}{2}\gamma a^2\delta^2$ (27). Where γ denotes the risk avoidance intensity of the hospital, determined by the risk awareness of the hospital's decision-making level. α indicates the incentive intensity, and δ represents the uncertain impact from random factors. Therefore, the actual investment cost of the hospital is $C_h = \frac{1}{2}\beta M^2 + \frac{1}{2}\gamma a^2\delta^2$. Where, $\delta \sim N(0, \sigma^2)$, indicates that the greater the variance of the random disturbance variable δ, the more the potential risk of the hospital is associated with the magnitude of its effort.

Benefit function of hospitals

After realizing the integration of EHR, not only can it bring economic benefits to hospitals, moreover, hospitals can obtain good social benefits. The economic benefits mainly include: first, the one-time subsidy given by the government to hospitals during the EHR interconnection stage; second, the incentive subsidy

given based on the performance of EHR sharing and use; and third, there is increased patient consumption, which results from the improved access efficiency and service quality due to EHR sharing. Social benefits mainly refer to good patient reputation, social image, and field status, which in turn are the foundation for improving hospital economic benefits. The potential social benefits of EHR integration include,: firstly, the public praise for reducing the financial burden of patients by reducing repeated examinations, which will attract more patients to seek medical treatment; secondly, the interconnection and interoperability of EHR can vigorously promote hierarchical diagnosis and treatment as well as upward and downward referrals, which will alleviate the heavy burden of large hospitals; and thirdly, the sharing and using EHR can enhance the accessibility of high-quality healthcare resources, and improve the overall regional healthcare conditions and environment for seeking medical treatment (28, 29). At the same time, it can strongly support hospitals to expand their existing business, such as providing Internet health consultation and health management.

The above benefits of hospitals is not only related to the profit level of the EHR (M), but also to the value-added coefficient of the EHR (τ), and to the coefficient of the hospital's ability to use the EHR (ρ). Therefore, the hospital's revenue can be expressed as $\pi_h(\rho,\tau) = \rho\tau M$, where $\tau \geq 0$ and $\rho \geq 0$. Interconnection does not add value to EHRs. The value added depends on the portion of EHR that are actually being used after sharing. Therefore, the value-added coefficient τ is actually the ratio of the number of EHR that are actually used to the number of all EHRs that are shared, and obviously $0 \leq \tau \leq 1$.

Incentive contract function

The government, to promote the EHR integration in medical consortiums, is bound to incentivize hospitals to actively participate in the sharing and use of EHRs. Generally, the government establishes incentive contracts either directly or by delegating to the medical consortiums. Assume that the one-time subsidy given by the government to hospitals in the EHR interconnection phase is H. The incentive contract function can be expressed as $S_h = (1-\alpha)H + \alpha\pi_h(\rho,\tau)$, where α is the incentive coefficient, $0 \leq \alpha \leq 1$. The incentive coefficients not only characterize the promotion strength for EHR integration across different consortiums or geographic regions, but also can accommodate trends in the evolution of incentive strength over time. When $\alpha = 0$, it means that the hospital does not bear the shared risk and does not receive the performance rewards; when $\alpha = 1$, it means that the hospital bears all the shared risk and receives the full performance rewards.

Basic incentive model

The above analysis shows that the expected benefits of the hospital are as follows:

$$\mathrm{EV_h} = (1-\alpha)H + \alpha\rho\tau M - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma\alpha^2\delta^2 \quad (1)$$

The expected benefits of the medical consortiums are as follows:

$$\mathrm{EV_c} = (1-\alpha)(\rho\tau M - H) \quad (2)$$

Therefore, the basic incentive model can be constructed as shown in Eq. (3):

$$\begin{cases} maxE(V) = \max\left[(1-\alpha)(\rho\tau M - H)\right] \\ s.t.\,(IR)\quad (1-\alpha)H + \alpha\rho\tau M - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma a^2\delta^2 \geq \overline{U} \\ (IC)\ \tau^* \in argmax\left[(1-\alpha)H + \alpha\rho\tau M - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma a^2\delta^2\right], \forall M^* \in A \end{cases} \quad (3)$$

Where, IR denotes the participation constraint, and IC represents the incentive constraint. $\overline{U}$ indicates the minimum utility before hospitals share EHR, which is the expected benefit of the hospital in the absence of incentive measures. A refers to the set of possible profit levels of the medical consortiums.

Incentive model of introducing performance evaluation construction

The basic incentive model allows hospitals to learn about each other's total shared EHR. However, the effort level hospitals can put into using shared EHR cannot be directly observed. To measure the hospital's effort level more accurately, it is necessary to design a supervisory signal in the incentive mechanism. Performance evaluation is the most commonly used supervision method in reality. After introducing a supervision signal using performance evaluation as a means, the final benefit of hospitals is not only related to the benefit level from using EHR, but also to the effectiveness of performance evaluation, thereby further improving the fairness of incentive contract.

Government and hospitals constitute a principal-agent relationship. The government can only indirectly judge and measure the efforts of hospitals through performance evaluation. Because there is a cost associated with conducting performance evaluations, which can be expressed in terms of kM. Where k denotes the effectiveness coefficient of performance evaluation. Larger k means that the results of performance evaluation are more realistic. For hospitals, performance evaluation may identify deficiencies in their shared use of EHRs, which will affect the amount of performance rewards. Performance evaluation effectiveness may be affected by random factors such as policy changes, optimization of evaluation criteria, information asymmetry. These potential stochastic effects are uniformly incorporated into δ to represent. With the inclusion of performance evaluation in the model, the one-time subsidy provided by the government will be affected by the results of the performance evaluation. Drawing inspiration from the research on supervision signals in the design of incentive mechanisms (30), when adding performance evaluation supervision signals to measure the hospital's effort level, the incentive model is as follows:

$$\begin{cases} maxE(V) = \max\left[(1-\alpha)(\rho\tau M - H) - kM\right] \\ s.t.\,(IR)\quad (1-\alpha)H + \alpha\rho\tau M + kM - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma(\alpha^2+k^2)\delta^2 \geq \overline{U_0} \\ (IC)\ \tau^* \in argmax\left[(1-\alpha)H + \alpha\rho\tau M + kM - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma(a^2+k^2)\delta^2\right], \forall M^{**} \in A \end{cases} \quad (4)$$

Where, *IR* denotes the participation constraint, and *IC* represents the incentive constraint. $\overline{U_0}$ denotes the retained utility of the hospital in the event of a performance evaluation. *A* is the set of possibilities for the hospital's level of effort.

Results and discussion

Solution and analysis of basic incentive model

By taking the first derivative of M for IC in (3) and setting the derivative value to 0, we get

$$M = \frac{\alpha\rho\tau}{\beta} \tag{5}$$

To ensure the enthusiasm of hospitals for sharing and using EHR, the benefits should not be less than the minimum utility before sharing and using EHR. Let *IR* in Eq. (3) take the equal sign and substitute $(1-\alpha)H$ into the objective function to transform the above optimization problem as follows:

$$maxE(V) = \rho\tau M - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma\alpha^2\delta^2 - \overline{U} \tag{6}$$

Taking the first order derivative of α with respect to Eq. (6), we get:

$$\alpha^* = \frac{\rho^2\tau^2}{\beta\gamma\delta^2 + \rho^2\tau^2} \tag{7}$$

Substituting α^* into Eq. (5), we get:

$$M^* = \frac{\rho^3\tau^3}{\beta^2\gamma\delta^2 + \beta\rho^2\tau^2} \tag{8}$$

As shown in Eqs. (7) and (8), both the level of hospital profit and the incentive coefficient are independent of the amount of one-time subsidy from the government. Therefore, one-time subsidies from the government are not the driving force to share and use EHRs in the long term, and cannot generate sustainable incentives. But it does not mean that the Government's one-time subsidies should be ignored because it is a safeguarding factor. When certain conditions are met, one-time subsidy from the government can help promote the smooth integration of EHR.

By taking partial derivatives of β, γ, ρ, τ and δ in Eq. (7), respectively, the relationship of the incentive coefficient α by a parameter can be obtained as follows:

$$\frac{\partial\alpha^*}{\partial\rho} = \frac{2\beta\gamma\delta^2\rho^2\tau^2}{(\beta\gamma\delta^2 + \rho^2\tau^2)^2} \geq 0$$
$$\frac{\partial\alpha^*}{\partial\tau} = \frac{2\beta\delta^2\rho^2\tau}{(\beta\gamma\delta^2 + \rho^2\tau^2)^2} \geq 0$$
$$\frac{\partial\alpha^*}{\partial\beta} = -\frac{\delta^2\rho^2\tau^2\gamma}{(\beta\gamma\delta^2 + \rho^2\tau^2)^2} \leq 0$$
$$\frac{\partial\alpha^*}{\partial\gamma} = -\frac{2\beta\delta^2\rho^2\tau^2}{(\beta\gamma\delta^2 + \rho^2\tau^2)^2} \leq 0$$
$$\frac{\partial\alpha^*}{\partial\delta} = -\frac{2\beta\delta\gamma\rho^2\tau^2}{(\beta\gamma\delta^2 + \rho^2\tau^2)^2} \leq 0$$

The above analysis shows that α^* is a decreasing function with respect to β, δ and γ and an increasing function with respect to ρ and τ. As the investment cost of hospitals increases, or as the risk avoidance intensity of hospitals increases and the risks brought by random disturbances increase, the government or medical consortium must also increase the incentive intensity to achieve the incentive purpose. With the continuous growth of hospitals' ability to share and use EHR, as well as the continuous increase in the value-added coefficient of EHR, the government can gradually reduce the incentive intensity appropriately, which will also fulfill the incentive purpose.

Solution and analysis of incentive models introducing performance evaluation

By taking the first derivative of *M* for IC in Eq. (4) and setting the derivative value to 0, we get

$$M'' = \frac{k + \alpha\rho\tau}{\beta} \tag{9}$$

Let *IR* in Eq. (4) take the equal sign and substitute $(1-\alpha)H$ into the objective function to transform the above optimization problem as follows:

$$maxE(V) = \rho\tau M - \frac{1}{2}\beta M^2 - \frac{1}{2}\gamma(a^2 + k^2)\delta^2 - \overline{U_0} \tag{10}$$

By taking the partial derivatives of α and k in Eq. (10), respectively, and making the derivative value zero, the following results can be obtained:

$$\alpha = \frac{\rho^2\tau^2 - \rho\tau k}{\beta\gamma\delta^2 + \rho^2\tau^2} \tag{11}$$
$$k = \frac{\rho\tau(1-\alpha)}{\beta\delta^2\gamma + 1} \tag{12}$$

Combining Equations (11) and (12) and solving the system of equations by the elimination method, we get:

$$\alpha^{**} = \frac{\rho^2 + \tau^2}{\beta\gamma\delta^2 + \rho^2\tau^2 + 1} \tag{13}$$
$$k^{**} = \frac{\rho\tau}{\beta\gamma\delta^2 + \rho^2\tau^2 + 1} \tag{14}$$

Substituting α^{**} and k^{**} into Eq. (9), we obtain:

$$M^{**} = \frac{\rho^3\tau^3 + \rho\tau}{\beta(\beta\gamma\delta^2 + \rho^2\tau^2 + 1)} \tag{15}$$

The effect of several parameters on the incentive coefficient α can be obtained by taking the partial derivatives of the variables in Eq. (13) separately, as follows:

$$\frac{\partial\alpha^{**}}{\partial\rho} = \frac{2\rho\tau^2(\beta\gamma\delta^2 + 1)}{(\beta\gamma\delta^2 + \rho^2\tau^2 + 1)^2} \geq 0$$

$$\frac{\partial \alpha^{**}}{\partial \tau} = \frac{2\rho^2\tau(\beta\gamma\delta^2+1)}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \geq 0$$
$$\frac{\partial \alpha^{**}}{\partial \gamma} = -\frac{\beta\delta^2\rho^2\tau^2}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \leq 0$$
$$\frac{\partial \alpha^{**}}{\partial \delta} = -\frac{2\beta\delta\gamma\rho^2\tau^2}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \leq 0$$
$$\frac{\partial \alpha^{**}}{\partial \beta} = -\frac{\delta^2\gamma\rho^2\tau^2}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \leq 0$$

The above analysis shows that the incentive coefficient α^{**}, which introduces performance evaluation, is a decreasing function with respect to β, δ, and γ and an increasing function with respect to ρ and τ. Therefore, the maximum incentive coefficient set by the government is positively related to the ability of hospitals to share and use EHRs and the value-added coefficient of EHR, while it is negatively related to the investment cost coefficient of hospitals, the intensity of hospitals' risk avoidance, and the intensity of the risk posed by random perturbations.

Similarly, the partial derivatives of Eq. (14) for each parameter can be obtained:

$$\frac{\partial k^{**}}{\partial \rho} = \frac{\tau(\beta\gamma\delta^2-\rho^2\tau^2+1)}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \geq 0$$
$$\frac{\partial k^{**}}{\partial \tau} = \frac{\rho(\beta\gamma\delta^2-\rho^2\tau^2+1)}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \geq 0$$
$$\frac{\partial k^{**}}{\partial \gamma} = -\frac{\beta\delta^2\rho\tau}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \leq 0$$
$$\frac{\partial k^{**}}{\partial \delta} = -\frac{2\beta\delta\gamma\rho\tau}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \leq 0$$
$$\frac{\partial k^{**}}{\partial \beta} = -\frac{\delta^2\gamma\rho\tau}{(\beta\gamma\delta^2+\rho^2\tau^2+1)^2} \leq 0$$

The above analysis shows that the performance evaluation effectiveness coefficient k^{**} is negatively related to β, δ and γ, and positively related to ρ and τ. The effectiveness coefficient of performance evaluation will increase with the ability of hospitals to share and use EHR, as well as the value-added coefficient of EHR. However, it will decrease with the increase of hospital investment cost coefficient, hospital risk avoidance intensity, and risk intensity caused by random disturbances.

Simulation and comparative analysis of incentive model

To verify the effectiveness of the incentive model, MATLAB was used for numerical simulation to visually compare the incentive effects before and after the introduction of performance evaluation and the impact of each key factor on the incentive mechanism. If the incentive coefficients and benefit levels after the inclusion of performance evaluation were greater than the previous coefficients and levels, provided that $\beta\gamma\delta^2-(1-\rho^2)\tau^2 \geq 0$ and $\beta^2\gamma\delta^2-(1-\beta)\rho^3\tau^3 \geq 0$ (Appendix A). Therefore, it is possible to $\beta = 0.7$, $\gamma = 0.6$, $\delta = 0.3$, $\tau = 0.2$, $\rho = 0.8$. When simulating the impact of a certain parameter on the incentive effect, the values of other parameters remain unchanged.

Impact of hospital investment cost coefficient β on incentive coefficients and benefit levels

As shown in Figure 1, the hospital investment cost coefficient β has a similar effect on the benefit levels before and after the inclusion of the performance evaluation monitoring signal, both of which gradually decrease as the hospital investment cost coefficient β increases. However, the incentive coefficients are more sensitive to the hospital investment cost coefficient β, which is not added to the performance evaluation. In the absence of a performance evaluation supervisory signal, the incentive effect is very good if the hospital investment cost coefficient is very low, and the incentive coefficient decreases rapidly as the hospital investment cost coefficient increases; when the hospital investment cost coefficient increases to 1, i.e., when the cost of EHR integration is fully borne by the hospital, the incentive coefficient becomes 0, indicating that the incentive will be lost altogether. Meanwhile, Figure 1 also indicates that the benefit level after adding performance evaluation is consistently higher than that without adding performance evaluation.

Impact of hospital risk avoidance intensity γ on incentive coefficients and benefit levels

As can be seen from Figure 2, both the incentive coefficient and the benefit level decrease gradually with the increase of hospital risk avoidance intensity, regardless of the inclusion of performance evaluation supervisory signals or not. The incentive coefficient is more sensitive to the hospital risk aversion intensity γ. When the performance evaluation supervisory signal is added, the strength of the hospital's risk avoidance intensity γ on both the incentive coefficient and the benefit level becomes smaller, suggesting that conducting performance evaluation is greatly beneficial for stabilizing the incentive effect. At this point, the impact of hospital risk avoidance intensity γ on the benefit level is consistently larger than when no performance evaluation supervisory signals are included, which may be mainly due to the additional costs associated with conducting performance evaluations.

Impact of capacity to use EHR ρ on incentive coefficients and benefit levels

As can be seen in Figure 3, both the incentive coefficients and the hospital benefit level will increase as the ability to use hospital records ρ increases, regardless of whether a performance evaluation supervisory signal is included in the incentive model. The impact of the hospital's ability to use EHR before adding performance evaluation supervision signal on incentive coefficients and benefit levels is smaller than after adding supervision signal. Conversely, after adding performance evaluation supervision signal, the hospital's ability to use EHR has a higher impact on improving incentive effectiveness and earnings level. It indicates that conducting performance evaluation has a

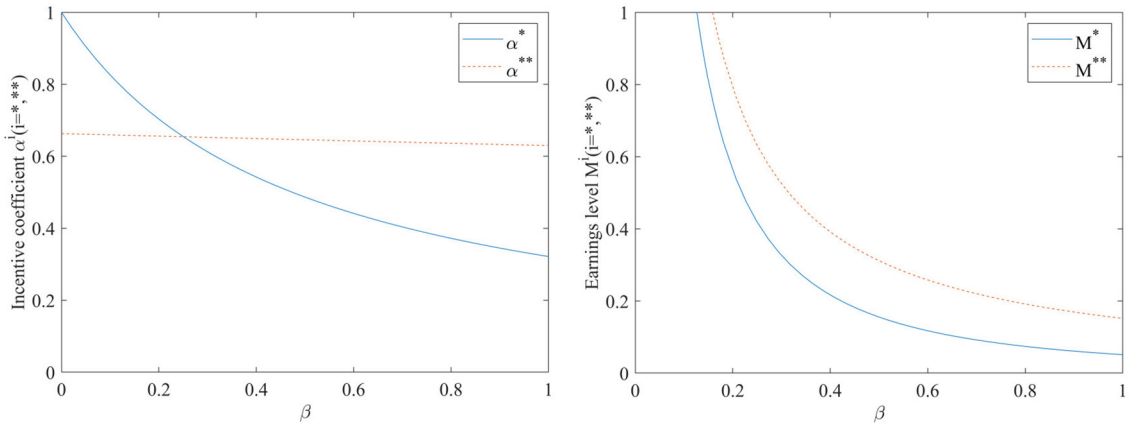

FIGURE 1
Impact of hospital investment cost coefficient β on incentive coefficients and benefit levels.

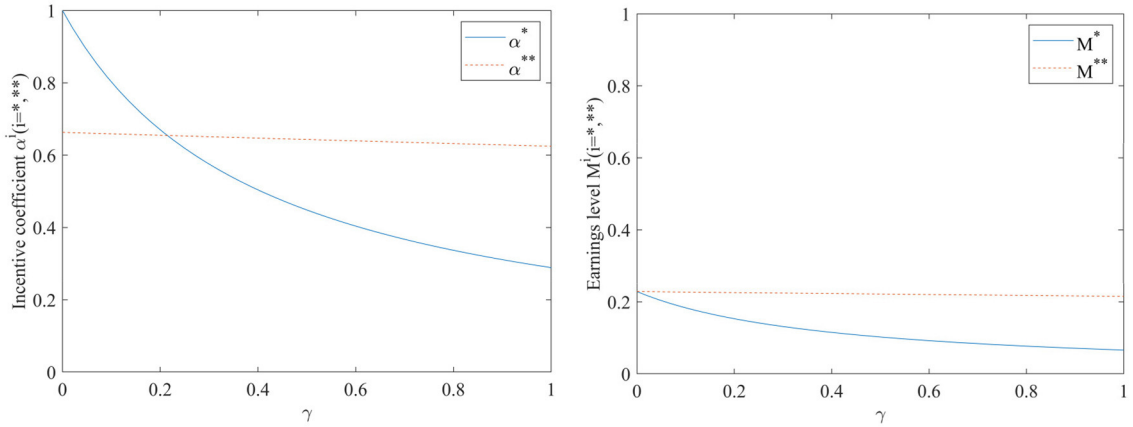

FIGURE 2
Impact of hospital risk avoidance intensity γ on incentive coefficients and benefit levels.

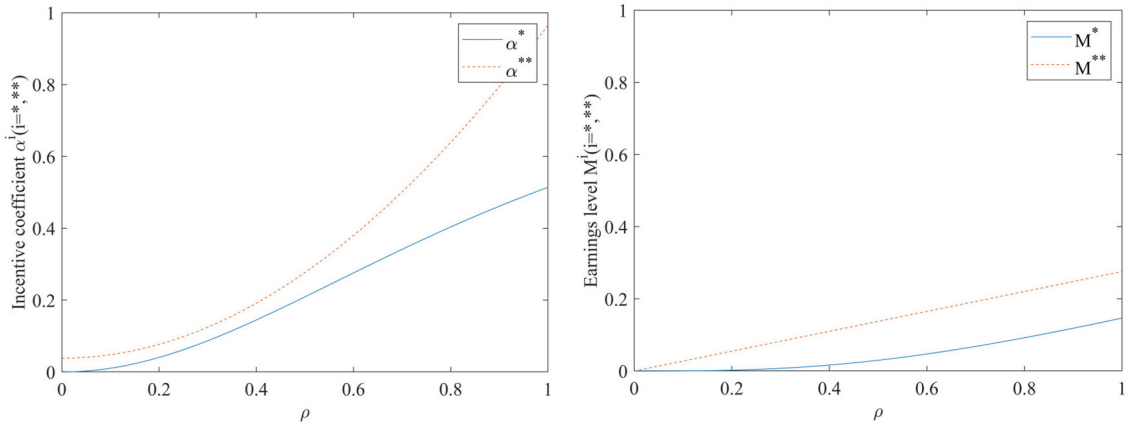

FIGURE 3
Impact of capacity to use HER ρ on incentive coefficients and benefit levels.

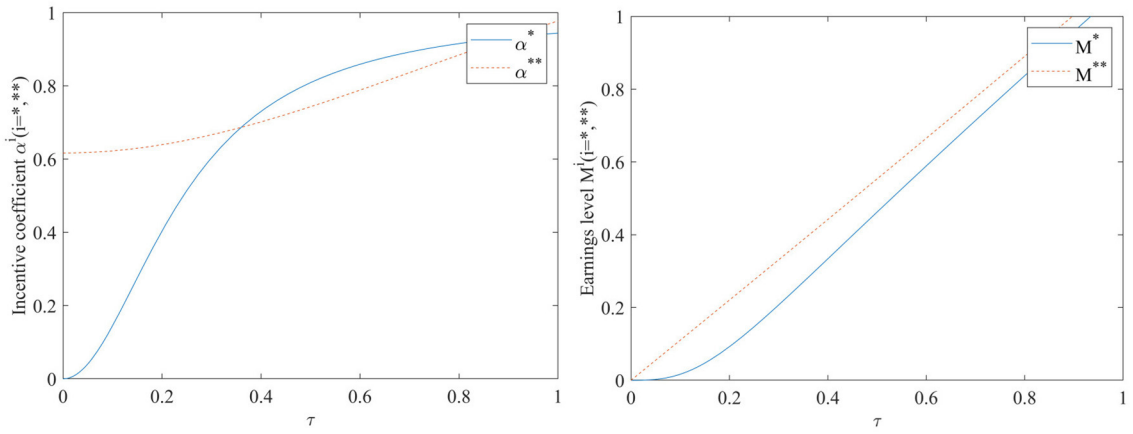

FIGURE 4
Impact of EHR value-added coefficient τ on incentive coefficients and benefit levels.

positive promoting effect on achieving sustainable sharing and use of EHR.

Impact of EHR value-added coefficient τ on incentive coefficients and benefit levels

As can be seen in Figure 4, the incentive coefficient and earnings level increase with the increase of value-added coefficient, regardless of whether or not the performance evaluation supervision signal is included in the model. When a performance evaluation supervision signal is added to the model, the incentive coefficient is at a high level even when the value-added coefficient is close to 0, and there is an almost linear increasing relationship between the value-added coefficient τ and the earnings level. In the case of adding performance evaluation supervision signal, the impact of the value-added coefficient on the earnings level is always greater than that of not adding performance evaluation supervision signal.

Impact of exogenous random disturbance factors δ on incentive coefficients and benefit levels

As can be seen in Figure 5, both the incentive coefficients and the earnings levels decrease as the exogenous random disturbance factors δ increases. When the performance evaluation supervision signal is not added, the influence of exogenous random disturbance factors on the incentive coefficient is very significant. As the exogenous random disturbance factors increase, the incentive coefficient will significantly decrease; At this point, the earnings level is also more sensitive to the impact of exogenous random disturbances. When the performance evaluation supervision signal is added, the effect of the exogenous random disturbance factors δ on both the incentive coefficients and the earnings level will become smoother. Figure 5 also shows that the earnings level with the inclusion of the performance evaluation supervision signal is consistently higher than that in the case of non-inclusion.

The effect of β, γ, ρ, τ, and δ on the performance evaluation effectiveness

To further analyze the effect of parameters such as β, γ, ρ, τ and δ on the effectiveness of performance evaluation, Figure 6 is plotted according to Eq. (14).

As can be seen from Figure 6, the value-added coefficient τ of the EHR has the greatest impact on performance evaluation effectiveness, and as the value-added coefficient increases performance evaluation effectiveness increases dramatically. The ability to use EHR ρ also has a significant positive effect on the effectiveness coefficient of performance evaluation. With the increase in the ability to use EHR the performance evaluation effectiveness coefficient also increases significantly. The effects of hospital investment cost coefficient β and hospital risk avoidance intensity γ on the performance evaluation effectiveness coefficient are basically the same, as they increase performance evaluation effectiveness coefficient slowly decreases. The effect of the exogenous random disturbance factors δ on the effectiveness coefficient of performance evaluation is slightly more pronounced than that of the intensity of risk avoidance γ, which also has a negative influence effect.

Conclusions and insights

In this paper, on the basis of constructing the basic incentive model, performance evaluation was introduced as the metric basis and supervisory signal of the hospital's efforts, and used numerical simulation methods to verify the effectiveness of incentive mechanism design. The findings of this paper not only clarify the motivational elements and their role relationships in EHR integration, but also provide a sustainable incentive scheme for achieving the goal of EHR integration, which will be an important reference value for further optimizing related policies and improving the promotion mechanism. The findings of this paper and its managerial implications mainly include the following aspects.

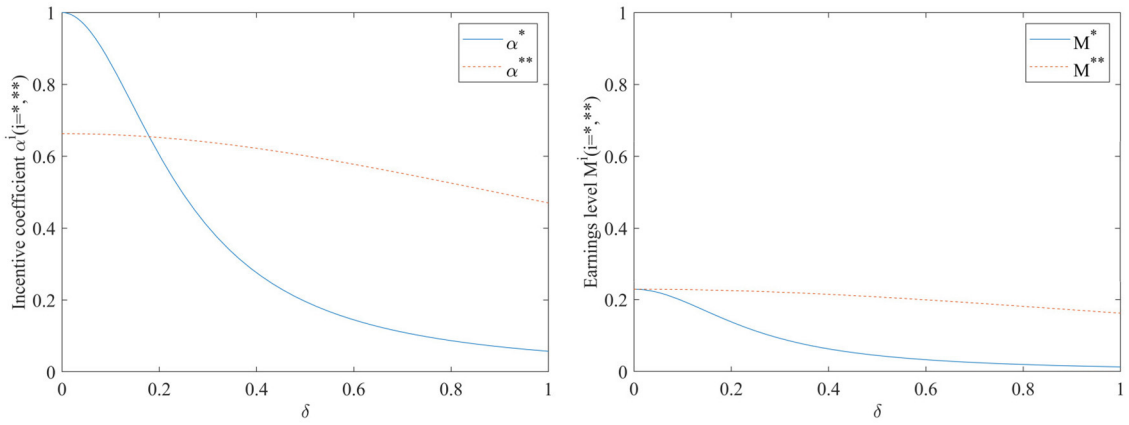

FIGURE 5
Impact of exogenous random disturbance factors δ on incentive coefficients and benefit levels.

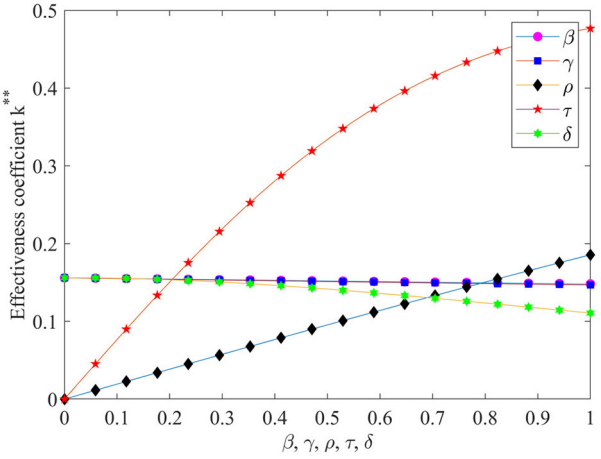

FIGURE 6
The effect of β, γ, ρ, τ and δ on the performance evaluation effectiveness. **It is used to distinguish between K *and K **.

Regardless of whether performance evaluation supervision signal is introduced or not, the incentive coefficient is an increasing function of the hospital's ability to use EHR and the EHR value-added coefficient, but a decreasing function of the hospital's investment cost, hospital risk avoidance intensity, and exogenous random disturbance factors. The ability to use EHRs of hospitals and the value-added coefficient of EHRs has a significant positive contribution to performance evaluation effectiveness. Therefore, seeking to enhance the capacity of hospitals to use EHR and to explore and expand the value-added space of EHR are 2 key paths to promote the sustainable sharing and use of EHR.

Neither the level of benefits nor the incentive coefficient for hospitals to share and use EHRs is related to the one-time government subsidy. The one-time government subsidy is a "hygiene factors" that is not a driving force for hospitals to share and use EHR for a long time, and cannot provide sustainable incentives. The government provides incentive subsidies based on the sharing and use performance of EHR, which is a more reasonable incentive method.

Conducting performance evaluation plays an important role in stabilizing the effectiveness of incentives. After incorporating the supervisory signal of performance evaluation into the model, the hospital's ability to use EHR has a higher impact on improving incentive effectiveness and earnings levels; The impact of exogenous random disturbance factors on incentive coefficients and earnings levels has also become more muted; The impact of the value-added coefficient on the earnings level is always greater than that without the addition of performance evaluation supervision signal. Therefore, effective performance evaluation is not only necessary but also important to advance the sustainable sharing and use of EHRs. The sharing and use of EHR should be included in the standardized maturity measurement programme for hospital information connectivity conducted annually in China to ensure objectivity and fairness in performance evaluation.

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

SHT: Formal analysis, Visualization, Writing – original draft. RJ: Formal analysis, Funding acquisition, Writing – original draft. YC: Methodology, Supervision, Writing – review & editing.

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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.

Publisher's note

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Appendix

Appendix A

(1) Analysis of changes in incentive coefficients before and after the introduction of performance evaluation

The incentive coefficient for adding performance evaluation supervision signal is $\alpha^{**} = \frac{\rho^2+\tau^2}{\beta\gamma\delta^2+\rho^2\tau^2+1}$, The incentive coefficient without the inclusion of the performance evaluation supervision signal is $\alpha^{*} = \frac{\rho^2\tau^2}{\beta\gamma\delta^2+\rho^2\tau^2}$. Therefore, there is

$$\alpha^0 = \alpha^{**} - \alpha^* = \frac{\rho^2+\tau^2}{\beta\gamma\delta^2+\rho^2\tau^2+1} - \frac{\rho^2\tau^2}{\beta\gamma\delta^2+\rho^2\tau^2} = \frac{\beta\gamma\delta^2-(1-\rho^2)\tau^2}{(\beta\gamma\delta^2+\rho^2\tau^2+1)(\beta\gamma\delta^2+\rho^2\tau^2)}$$

To make $\alpha^0 \geq 0$ means that in order for performance evaluation to be effective, it is necessary to $\beta\gamma\delta^2-(1-\rho^2)\tau^2 \geq 0$.

(2) Analysis of changes in the earnings level before and after the introduction of performance evaluation

The benefit level included in the performance evaluation supervision signal is $M^{**} = \frac{\rho^3\tau^3+\rho\tau}{\beta(\beta\gamma\delta^2+\rho^2\tau^2+1)}$, The benefit level without adding performance evaluation supervision signals is $M^{*} = \frac{\rho^3\tau^3}{\beta^2\gamma\delta^2+\beta\rho^2\tau^2}$. Therefore, it can be obtained that:

$$M^0 = M^{**} - M^* = \frac{\beta^2\gamma\delta^2+(\beta-1)\rho^3\tau^3}{\beta(\beta\gamma\delta^2+\rho^2\tau^2+1)(\beta^2\gamma\delta^2+\beta\rho^2\tau^2)}$$

To make $M^0 \geq 0$, which means that the earnings level after adding performance evaluation increases, it is necessary to meet the following requirements: $\beta^2\gamma\delta^2-(1-\beta)\rho^3\tau^3 \geq 0$.

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Cross-sectional comparison of health care delivery and reimbursement between segregated and nonsegregated communities in Hungary

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Introduction: Spatially segregated, socio-economically deprived communities in Europe are at risk of being neglected in terms of health care. In Hungary, poor monitoring systems and poor knowledge on the health status of people in these segregated areas prevent the development of well-informed effective interventions for these vulnerable communities.

Aims: We used data available from National Health Insurance Fund Management to better describe health care performance in segregated communities and to develop more robust monitoring systems.

Methods: A cross-sectional study using 2020 health care data was conducted on each general medical practice (GMP) in Hungary providing care to both segregated and nonsegregated (complementary) adult patients. Segregated areas were mapped and ascertained by a governmental decree that defines them as within settlement clusters of adults with low level of education and income. Age, sex, and eligibility for exemption certificate standardized indicators for health care delivery, reimbursement, and premature mortality were computed for segregated and nonsegregated groups of adults and aggregated at the country level. The ratio of segregation and nonsegregation specific indicators (relative risk, RR) was computed with the corresponding 95% confidence intervals (95% CI).

Results: Broad variations between GMPs were detected for each indicator. Segregated groups had a significantly higher rate of health care service use than complementary groups (RR = 1.22, 95% CI: 1.219;1.223) while suffering from significantly reduced health care reimbursement (RR = 0.940, 95% CI: 0.929;0.951). The risk of premature mortality was significantly higher among segregated patients (RR = 1.184, 95% CI: 1.087;1.289). Altogether, living in a segregated area led to an increase in visits to health care services by 18.1% with 6.6% less health spending.

Conclusion: Adults living in segregated areas use health care services more frequently than those living in nonsegregated areas; however, the amount of health care reimbursement they receive is significantly lower, suggesting lower quality

of care. The health status of segregated adults is remarkably lower, as evidenced by their higher premature mortality rate. These findings demonstrate the need for intervention in this vulnerable group. Because our study reveals serious variation across GMPs, segregation-specific monitoring is necessary to support programs sensitive to local issues and establish necessary benchmarks.

KEYWORDS

cross-sectional, segregation, inequality, healthcare, health reimbursement, general medical practitioner, Hungary

1 Introduction

Segregation, whether residential, racial, or otherwise, is a recognized risk factor for ill health and inequity. Various studies have explored segregation and its impact on health (1–7) and found pronounced inequality in health care outcomes between segregated and nonsegregated areas, including but not limited to overall infant and adult mortality, high-risk pregnancies, and exposure to communicable diseases (7, 8). Reasons for these disparities range from environmental factors, such as sanitation facilities and pollution, to lifestyle factors, such as poor housing situations, crowding, and habits such as poor diets, smoking, and low physical activity (9, 10).

The relationship between segregation and health is well documented. However, the specific association of segregation with health care delivery and reimbursement policies is still understudied. A prominent example of this is the Roma people in Europe, who constitute the largest ethnic minority in Europe (11, 12) and in Hungary (13), where they make up 94% of inhabitants in segregated areas (14). Data regarding their health are limited. However, it is estimated that Roma have a 10-year shorter life expectancy (15) and are at higher risk of coronary and chronic diseases (16, 17). It makes them among the most relevant health equity concerns in Europe and prompts many studies to investigate segregated Roma access to and use/misuse of health care services (18–22). However, these studies have fallen short of explaining the determinants of the observed health losses. This shortfall has contributed to the failure of governments to implement effective equity-targeted policies, as evidenced by the current dissimilarity in health between Roma and non-Roma populations (23).

A major constraint of any Roma-focused study is the inability to accurately identify Roma ethnicities in local demographics or estimate their health statistics, mainly due to ethical issues and the unavailability of a clear-cut method to do so (24–26). These limitations generate unguided financial policies and governance, leading to considerable inequality in health delivery and outcomes both geographically and among population groups (27, 28). This situation gives rise to the need for more robust monitoring and intervention programs, both of which require a clearer picture of the variability in health care delivery and reimbursement.

Moreover, even though Roma are overrepresented in segregated communities in Hungary, the non-Roma population of the segregates lives in equal socioeconomic deprivation, and interventions should therefore be sensitive to this situation and the local environment and aim to help the vulnerable population regardless of ethnic background.

As a result, current inclusion policies shift the focus from Roma people to segregated communities regardless of ethnicity, to avoid any ethical concerns surrounding the identification of different ethnic groups as well as monitoring their health status, in accordance with the Hungarian national social inclusion strategies (29).

In Hungary, the National Institute of Health Insurance Fund Management (NIHIFM) is the organization operating the country's official health monitoring system. Every month, NIHIFM evaluates all general medical practices (GMPs) by a limited set of performance indicators (30, 31), which then affect GMP financing through a pay-for-performance system. More data on primary health care operations are collected but not utilized by the NIHIFM, leaving an untapped reservoir of data that could be useful for research.

Using the protected data within the NIHIFM system, aggregated indicators can be produced for segregated areas where most inhabitants are Roma and other vulnerable groups that are cared for by identifiable institutions, achieved by linking the geographical location of segregation with the health-insured population living there. These aggregated statistics can then be used to investigate the most vulnerable subgroup of Roma by evaluating geographical inequality (32, 33), thus bypassing legal and ethical limitations. The governmental decree's definition of segregation utilizes census data, focusing on income and education measures rather than Roma ethnicity, which enables the study of segregation related issues and leads to conclusions indirectly related to Roma.

The purpose of our study was (1) to use the available but untapped NIHIFM data as a means to describe patient inequality in health delivery and reimbursement between segregated communities (where the most vulnerable populations with extremely high Roma representation reside) and nearby nonsegregated areas; (2) to outline the variability of this inequality across different Hungarian GMPs that serve both segregated and nonsegregated areas; and (3) to aid in the conceptualization and implementation of a new equality-oriented monitoring system.

2 Methods

2.1 Setting

The study utilized person-level health records, and evaluated GMP-level-aggregated indicators. All Hungarian GMPs ($N = 4,359$)

Abbreviations: GMP, General medical practice; GP, General practitioner; NHIF, National Health Insurance Fund; NIHIFM, National Institute of Health Insurance Fund Management; SA, Segregated areas; CA, Complementary areas; CI, Confidence interval; IQR, Inter quartile range; CT, Computed tomography; MRI, Magnetic resonance imaging.

who delivered care for adults were investigated. Each of them were contracted with the NIHIFM (the only health insurance institute in Hungary). The NIHIFM provided data on 2020 for secondary analysis on the health care use, reimbursement, and health status of adults belonging to the GMPs.

2.2 Design

A cross-sectional study of Hungarian GMPs providing care to segregated adults was performed. Segregated areas (SAs) were mapped and ascertained by applying the classification of a governmental decree that defines them as within settlement clusters of adults between the ages of 18 and 59 with a high proportion with at most primary-level education and no active income. Accordingly, the NIHIFM classified each household as either an SA or nonsegregated area [complementary area (CA)], which were mutually exclusive. Using addresses, any adult of at least 18 years of age could therefore be defined as living in an SA or *CA*. GMPs without patients living in an SA were excluded from the analysis.

2.2.1 Outcome indicators

2.2.1.1 Health care delivery

Health care delivery rates for multiple services were calculated as the number of patients who used the health service per patient belonging to a GMP for the previous 12 months. The delivery indicators included (1) the number of general practitioner visits, (2) outpatient service use without computed tomography or magnetic resonance imaging (CT/MRI) services, (3) CT/MRI services, and (4) hospitalization.

2.2.1.2 Health care reimbursement

Per capita reimbursement was calculated for these services as health insurance spending (in Euros) per patient belonging to the general medical practice for the previous 12 months. With the addition of medication costs, and since the NIHIFM finances GMPs *per capita* irrespective of the number of patient visits, this measure was not included among the reimbursement indicators, as it does not influence variability in GMP average *per capita* financing.

2.2.1.3 Premature mortality

All-cause premature mortality was defined as all deaths of adults below 65 years of age who had not changed their GMP in the past 5 years. This restriction was applied to exclude those who died in the care of a new GMP who did not manage their health prior to their death.

2.3 Statistical analysis

Standardized performance measures were calculated using the national average as a reference. Indicators for SAs and CAs were indirectly standardized by age (applied age groups: 18–24, 25–29, 30–34, 35–39, 40–44, 45–49, 50–54, 55–59, 60–64, 65–69, 70–74, 75–79, and 80 and above), sex and, eligibility for an exemption certificate. Exemption certificates are issued by the local municipalities to patients with disadvantageous socioeconomic status and chronic diseases (if recommended by GPs) and ensure free of charge access to medicines and medical devices. If GMPs provided care to more than one settlement, the observed and expected values were summarized to obtain GMP specific SA and CA measures. Dividing GMP-level observed values over expected values resulted in standardized risk ratios for SAs (SRsa) and CAs (SRca) for each indicator and GMP (32, 34–36). These GMP-level data were aggregated further to obtain country level standardized measures for SAs and CAs.

Relative performance in SAs was described by the risk ratio (RR), which was calculated by the SRsa/SRca ratio for each GMP and for the whole country along with 95% confidence intervals (95% CIs). Impact measures such as excess number of cases in the SAs, percentage of risk attributable to segregation in the population of SAs (attributable risk), and percentage of risk attributable to segregation in the population of the entire country (population attributable risk) were computed using nationally and locally adjusted standardized ratios.

3 Results

The studied population consisted of 7,385,641 adults (3,456,560 men and 3,929,081 women), with 2,071 identified segregated areas hosting 283,876 adults (139,507 men; 144,369 women). Demographic structure varied widely between the SA and CA populations (Figure 1). The mean age was notably lower among those living in SAs (total: 43.3 years; men: 42.2 years; women: 44.4 years) than among those living in CAs (total: 50.4 years; men: 48.5 years; women: 52.1 years). The older adult dependency ratio (the proportion of older adult individuals aged 65 or above among individuals aged 15 and above) was remarkably lower in SAs (15.4%) than in CAs (33.7%).

The distribution of the number of segregated patients belonging to a GMP was highly varied across the standardized health care delivery, health care reimbursement, and premature mortality indicators (Figures 2–4). The highest level of heterogeneity of both episode and reimbursement indicators for relative GMP performance was observed in imaging examinations. Outpatient service use showed the highest variability.

3.1 Health care delivery

According to the standardized relative indicators aggregated for the whole country (Table 1) segregated groups had a significantly higher rate of health care services use than their complementary counterparts (RR = 1.222, 95% CI: 1.220;1.223). Specifically, for individual services provided to segregated patients, the number of GP visits per person per year (RR = 1.251, 95% CI: 1.249;1.253) and the number of hospital admissions per year exceeded those for complementary patients (RR = 1.250, 95% CI: 1.237;1.264). on the other hand, segregated patients experienced a significantly reduced number of treatments per year in outpatient service centers (RR = 0.948, 95% CI: 0.943;0.953) and imaging examinations (RR = 0.935, 95% CI: 0.920;0.950) compared to their counterparts.

Regarding attributable risk (Table 2), patients living in an SA were associated with an increase in visits to health care services by 18.1% (95% CI: 18.0;18.2), with a population-attributable risk of 0.742% when comparing with CAs. Considering the number of episodes, the number of GP visits showed broad inequality, with an estimation of 400,024 (95% CI: 397,811;402,234) excess visits made per year. Hospital service use also had an excess of 7,116 (95% CI: 6,819;7,410),

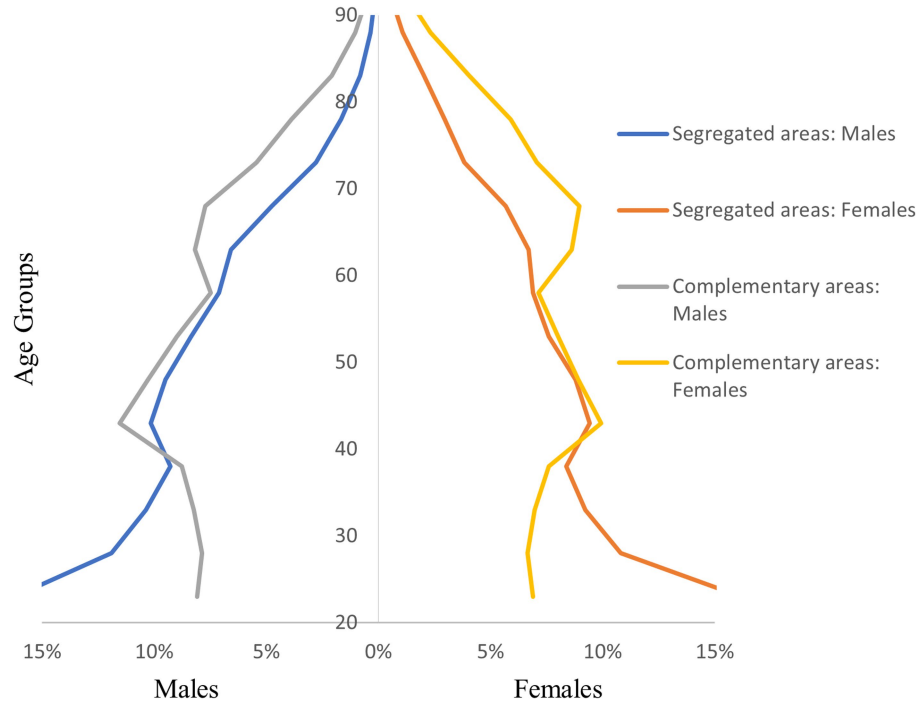

FIGURE 1
Demographic structure of the segregated and complementary populations.

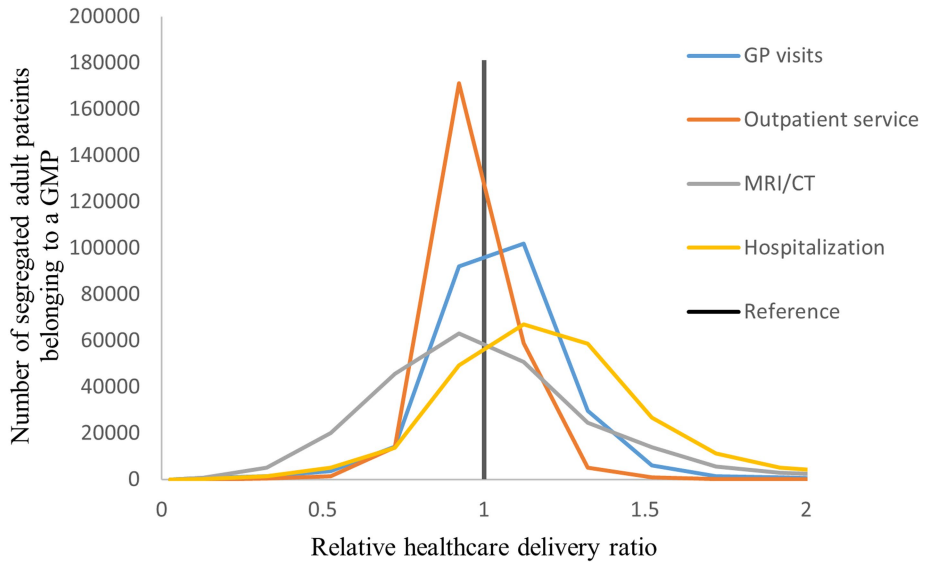

FIGURE 2
Distribution of segregated adult patients according to the relative health care delivery ratio among Hungarian GMPs.

a 20% increase compared to residence in a *CA*. Meanwhile, the number of outpatient services used and CT/MRI examinations were lower by 8,241 (95% CI: 9,045;7,441) and 1,046 (95% CI: 7,911;306), respectively.

3.2 Health care reimbursement

Health care reimbursement also had significant dissimilarities depending on whether patients lived in SAs or CAs (Table 1). Total health services reimbursement for a GMP was significantly reduced for SA patients (RR = 0.938, 95% CI: 0.927;0.950). Specifically, GMPs exhibited significantly lower reimbursement per year for outpatient services to SA patients (RR = 0.878, 95% CI: 0.848;0.908), MRI/CT examinations (RR = 0.815, 95% CI: 0.752;0.883), and medications (RR = 0.869, 95% CI: 0.854;0.884). Hospitalization reimbursement, on the other hand, showed higher spending on segregated groups (RR = 1.063, 95% CI: 1.043;1.083).

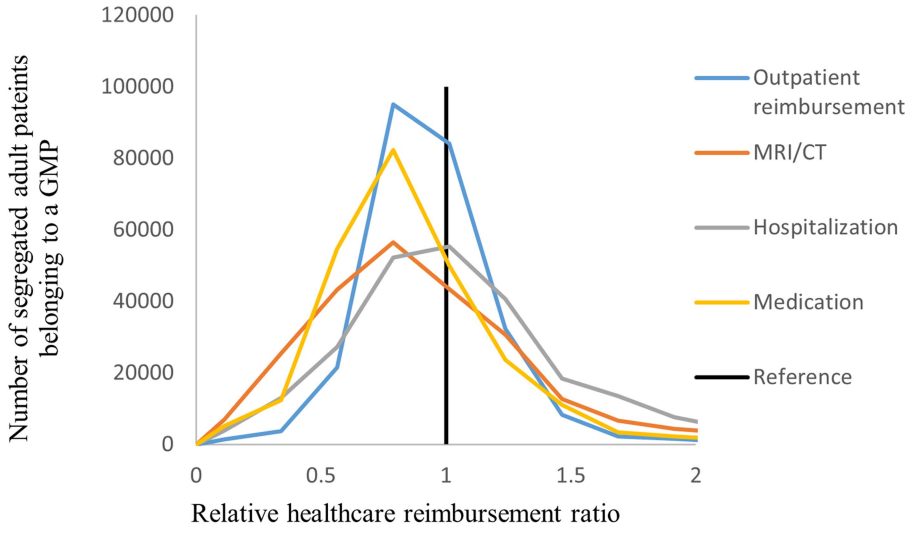

FIGURE 3
Distribution of segregated adult patients according to the relative health care reimbursement ratio among Hungarian GMPs.

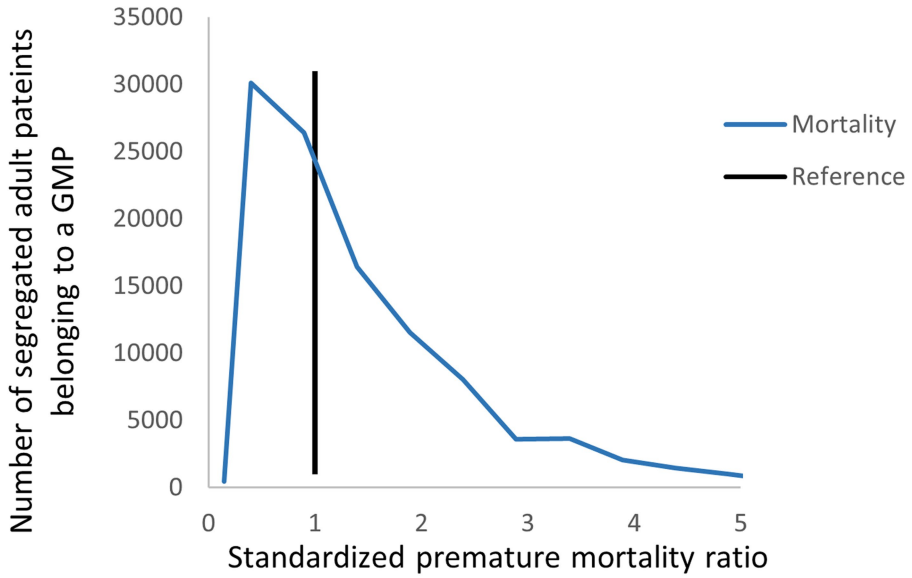

FIGURE 4
Distribution of segregated adult patients according to the relative premature mortality ratio among Hungarian GMPs.

Providing care to segregated patients lowered health care reimbursement by 6.6% (95% CI: 7.8;5.3) (Table 2) when compared to complementary groups, with a population-attributable risk of −0.213%. This finding is predominantly attributed to medication-related NIHIFM reimbursement, where the *per capita* yearly cost was lower for SA patients by 1,899 EUROs (95% CI: 2,154;1,648) compared to CA patients.

3.3 All-cause premature mortality

The age-and sex-standardized premature mortality among the SA population was significantly higher than that in the CA population (RR = 1.092, 95% CI: 1.030;1.157) (Table 1). The estimated 101.544 (95% CI: 37.355;162.213) excess cases in the SA population corresponded to 8.406% (95% CI: 3.092;13.428). The population level impact was estimated to be 0.433%.

4 Discussion

4.1 Main findings

Our study reveals variation and statistically significant dissimilarity in health care use, reimbursement, and premature mortality across Hungarian GMPs, pointing to causes of the existing health gap between segregated people and their counterparts living in complementary areas.

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TABLE 1 Relative health care delivery, reimbursement, and all-cause premature mortality among Hungarian adults provided by general medical practices situated in a segregated or complementary area.

Indicators	Total	Segregated colonies		Complementary areas		Relative performance in segregated colonies [95%CI#]
	N	*N*	Standardized performance* [95%CI#]	*N*	Standardized* performance [95%CI#]	
Healthcare delivery (episodes)						
GP visits	47,754,032	1,993,344	1.238 [1.237;1.240]	45,760,688	0.990 [0.989;0.990]	1.251 [1.249;1.253]
Use of outpatient services without CT/MRI	4,522,976	150,414	0.951 [0.946;0.956]	4,372,562	1.003 [1.002;1.004]	0.948 [0.943;0.953]
Use of CT/MRI	493,566	15,078	0.940 [0.925;0.955]	478,488	1.005 [1.002;1.008]	0.935 [0.920;0.950]
Use of hospital service	836,818	35,527	1.241 [1.228;1.254]	801,291	0.992 [0.990;0.994]	1.250 [1.237;1.264]
Total	53,607,392	2,194,363	1.211 [1.209;1.212]	51,413,029	0.991 [0.990;0.991]	1.222 [1.220;1.223]
Healthcare reimbursement (Euro *per capita*)						
Use of outpatient services without CT/MRI	42.67	35.31	0.885 [0.856;0.915]	42.94	1.008 [1.002;1.014]	0.878 [0.848;0.908]
Use of CT/MRI	8.57	6.34	0.823 [0.760;0.890]	8.65	1.009 [0.996;1.022]	0.815 [0.752;0.883]
Hospital service	124.63	117.36	1.062 [1.043;1.082]	124.91	0.999 [0.996;1.003]	1.063 [1.043;1.083]
Use of hospital service	147.18	130.01	0.871 [0.856;0.887]	147.82	1.003 [0.999;1.006]	0.869 [0.854;0.884]
Total	323.05	289.02	0.940 [0.929;0.952]	324.33	1.002 [1.000;1.004]	0.938 [0.927;0.950]
All cause premature mortality						
All-cause premature mortality	23,453	1,208	1.087 [1.027;1.150]	22,245	0.996 [0.983;1.009]	1.092 [1.030;1.157]

*Age, sex, and eligibility for exemption certificate standardized. #95% confidence interval.

A characteristic health care use pattern of adults living in segregated areas was identified. People living in SAs use health care services more frequently than those living in CAs; however, the amount of health care reimbursement paid for their care is significantly lower, suggesting lower quality of care.

In the case of primary care, crude indicators show that segregated groups had a higher rate of GP appointments, which corresponds with other studies from Europe (37, 38). And publications on the poorer health status of residents of segregations (32, 35, 36).

However, the services of outpatient and imaging centers were notably less utilized by segregated groups, which correlates with a significant reduction in GMPs' reimbursement compared to complementary groups. This difference, coupled with the fact that people living in SAs have poorer health status and a likely greater need for outpatient services, implies the underutilization of these services by segregated groups.

This finding helps explain the increase in hospital service utilization in SA groups paralleled with an increase in reimbursement; since outpatient services (including advanced and specialized tests and treatments) are required to diagnose, reverse, or halt the progression of chronic diseases, which can otherwise go undetected until a more serious prognosis requires hospitalization. Moreover, this discrepancy could also be due to the poorer health and greater needs of people living in SAs. Numerous studies have reported increased hospital admissions, avoidable and otherwise, in segregated minorities (32, 39, 40).

The health status of adults living in segregation was significantly poorer than that of adults living in complementary areas, as evidenced by their higher premature mortality rate, corroborating other studies showing the same conclusion (17, 34, 35). Their poorer health may be an outcome of their lower socioeconomic status and unhealthy lifestyles (10, 41) combined with the provision of lower-quality health care (30).

4.2 Strengths and limitations

The major strength of our study was the inclusion of all Hungarian adults living in SAs or in CAs. Because the Hungarian GMPs are required to contract with NIHIFM, detailed reimbursement and health data were available for each subject. Therefore, there was no selection bias in our study. Further, the proper quality of data was ensured by the standard NIHIFM protocols for data collection (42).

The present study results reflect estimations of health care use and mortality of Roma living in SAs, since Roma constitute 94% of the inhabitants of these areas (14). It is presumable that the Roma and non-Roma adults in SAs follow similar lifestyle and have similar health

TABLE 2 Impact of segregation among adults living in segregated areas (number of excess cases and attributable risk) and in the whole adult population of Hungary (population attributable risk).

Indicators	Number of excess cases [95%CI#]	Attributable risk [95%CI#]	Population attributable risk
Healthcare delivery (episodes)			
GP visits	400,024 [397,811;402,234]	20.1% [20.0%;20.2%]	0.838%
Use of outpatient services without CT/MRI	−8,241 [−9,045;−7,441]	−5.5% [−6.0%;−4.9%]	−0.182%
Use of CT/MRI	−1,046 [−1,306;−791]	−6.9% [−8.7%;−5.2%]	−0.212%
Use of hospital service	7,116 [6,819;7,410]	20.0% [19.2%;20.9%]	0.850%
Total	397,921 [395,543;400,297]	18.1% [18.0%;18.2%]	0.742%
Healthcare reimbursement (Euro *per capita*)			
Use of outpatient services without CT/MRI	−477 [−609;−348]	−14.0% [−17.8%;−10.2%]	−0.418%
Use of CT/MRI	−139 [−201;−81]	−22.7% [−32.8%;−13.3%]	−0.607%
Hospital service	674 [476;869]	5.9% [4.2%;7.7%]	0.203%
Medication	−1,899 [−2,154;−1,648]	−15.1% [−17.1%;−13.1%]	−0.483%
Total	−1,838 [−2,190;−1,491]	−6.6% [−7.8%;−5.3%]	−0.213%

status. Consequently, the SA specific indicators can be considered as proxy estimations for those Roma who live in segregated areas.

Unfortunately, the health care use indicators of our study were not adjusted for the health needs and health status of the investigated populations, to which the observed differences in health care use could be partially attributed. The higher premature mortality shows that the health status is worse in SAs, consequently leading to more intensive use of health care services in these areas. In this context, the higher frequency of GP visits and hospital admissions can be considered an adequate response to the higher health care needs, while the lower utilization of outpatient services in SAs is inconsistent, suggesting a potential malfunction in the system, where fewer outpatient services were provided than necessary. Our results do not identify the mechanisms behind the observed inequalities, calling for more detailed pathway analyses to propose interventions that could help lessen the present inequality.

Data available for our analysis were from 2020. That year was seriously affected by COVID-19. The first case in Hungary was detected on 04/03/2020 (43). The consecutive epidemiologic measures profoundly impacted health care operation (44). Consequently, our results predominantly reflect health care inequality during the COVID-19 epidemic. However, the compared groups of SAs and CAs shared the same settlements, geographical access to health care, and GP who was ultimately responsible for the gate-keeping of treatment pathways. Therefore, any differences between SAs and CAs in health care use or reimbursement should be minimized. Nevertheless, vaccination coverage was lower in SAs, suggesting that the COVID-19 epidemic contributed to some of the observed health care use and reimbursement inequalities (45). Altogether, the inequality pattern observed in our study should be further investigated in years not affected by the epidemic to substantiate the findings of our investigation.

4.3 Implications

Our findings indicate a significant association between segregation and severe health care issues, as demonstrated by country level aggregated relative risk measures. Notably, these problems largely stem from the local setting as evidenced by the varying levels of inequality at the local GMP-level. Some GMPs exhibit pronounced local inequalities, while others show no such disparities (Supplementary Table S1).

A monitoring system could distinguish between GMPs with and without local bias, and could monitor the time trend of country level inequality. At present, there is no monitoring system, to inform stakeholders neither at local nor at the country level. Consequently, there is a pressing need for monitoring systems specific to SAs. Our investigation demonstrates not only the feasibility of segregation oriented monitoring but also suggests indicators for this purpose. However, it is evident that additional indicators are required to understand the processes leading to the observed disparities in premature mortality, health care use, and insurance reimbursement. This monitoring system could support the National Social Inclusion Strategy of the Hungarian government (29), and as Roma are overrepresented in SAs, it could contribute to the programs aimed at improving the health status of the Roma population. The effectiveness of interventions could be enhanced by considering self-declared Roma ethnicity (available from census data) in defining SAs.

5 Conclusion

This analysis showed that (1) in the Hungarian health system, there are varying degrees of GMP-level dissimilarity in both health services delivery and reimbursement, in addition to varying health status between people living in SAs and CAs; this suggests that residence in an SA is a strong factor impacting the health care services system. Furthermore, (2) some Hungarian GMPs seemed to provide equal care to their inhabitants, while others show varied levels of inequality. We suggest that further studies are required to investigate such variations and local factors affecting the quality of care provided to segregated populations. According to our findings (3) any decision-making on interventions related to SAs should take the local (GMP-level) environment into consideration.

Data availability statement

The data analyzed in this study is subject to the following licenses/restrictions: the datasets used and/or analyzed during the current study are available from the corresponding author on reasonable

request. Requests to access these datasets should be directed to janos.sandor@med.unideb.hu.

Ethics statement

The protocol to produce segregation-specific indicators was approved by the Office of the Commissioner for Fundamental Rights (AJB-3147/2013), the general director of the NHIF (E0101/215-3/2014), and the Hungarian National Authority for Data Protection and Freedom of Information (NAIH/2015/826/7N). The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

FK prepared the literature review, analyzed, interpreted the data, and prepared the draft of the manuscript. FV prepared the primary database. LK helped with conception and data base preparation. RÁ helped with conception and study design. JS elaborated on the design, interpreted the results, and finalized the manuscript. All authors read and approved the final manuscript.

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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.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1152555/full#supplementary-material

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The socioeconomic burden of spinal muscular atrophy in Saudi Arabia: a cross-sectional pilot study

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Background: Spinal muscular atrophy (SMA) is a rare debilitating condition with a significant burden for patients and society. However, little is known about how it affects Saudi Arabia's population. The socioeconomic and medical characteristics of affected SMA patients and their caregivers are lacking.

Purpose: This study aimed to describe the socioeconomic and medical characteristics of SMA patients and caregivers in Saudi Arabia.

Patients and methods: A cross-sectional questionnaire-based study was conducted using snowball sampling. Assessment tools including EuroQol (EQ-5D-5L) and visual analog scale (EQ-VAS), Generalized Anxiety Disorder 7-item (GAD-7), Patient Health Questionnaire (PHQ-9), and Costs for Patients Questionnaire (CoPaQ) were used to assess the quality of life (QoL), anxiety, depression, and out-of-pocket expenditures.

Results: Sixty-four caregivers of SMA patients participated. Type I patients had higher sibling concordance, ICU hospitalization, and mechanical support needs. Type III patients had better QoL. Type I patients' caregivers had higher depression scores. Type III patients' caregivers had higher out-of-pocket expenditures. Forty-eight percent received supportive care, while others received SMA approved therapies.

Conclusion: SMA imposes a significant socioeconomic burden on patients and caregivers, requiring more attention from the healthcare system. Access to innovative therapies varied across SMA types. Pre-marital screening and early detection are crucial to reduce disease incidence and ensure timely treatment.

KEYWORDS

access to treatments, burden of disease, Saudi Arabia, spinal muscular atrophy, out-of-pocket (OOP) expenses

1 Introduction

Spinal Muscular Atrophy (SMA) is a hereditary neurodegenerative disease that primarily affects nerve cells in the anterior horn of the spinal cord, leading to irreversible degradation of α-motor neurons within the anterior horn cells and brain stem nuclei (1, 2). This genetically-linked neuromuscular condition significantly impacts the musculature of the upper limbs, reducing working capacity and ultimately causing respiratory distress due to diaphragm involvement, placing considerable burden on caregivers and decreasing the patient's life expectancy (3, 4).

Approximately 95% of SMA cases arise from homozygous deletions or mutations in survival motor neuron 1 (SMN1) on chromosome 5q13, leading to a decrease in SMN protein expression (5). However, the SMN2 gene serves as a compensatory mechanism, although only about 10–20% of SMN2-expressed protein is fully functional (1). Therefore, increased SMN protein copy numbers may alleviate disease severity (6).

SMA has diverse clinical presentations, divided into five types based on disease progression and symptom onset, with varying impacts on life expectancy (7). Furthermore, its manifestation is often categorized into four phenotypes according to motor function and age of onset (8). The Werdnig-Hoffmann variant (Type I) is the most prevalent and severe diagnosed within the first 6 months of life, is little bit less progressive and diagnosed early in the childhood (e.g., between 6 and 18 months), the Kugelberg-Welander (Type III) is the mildest and is diagnosed after the child's first 18 months, while type IV is a rare and mild type of the disease and its symptoms mostly appear in the mid-thirties (6). Symptomology often includes symmetrical muscle weakness, respiratory complications, and paralysis in severe cases (3, 9).

The management of SMA primarily focuses on supportive care, encompassing provision of sufficient nourishment, respiratory support, and mitigation of muscular weakness effects through therapeutic interventions or preventative measures (10). This includes hospitalizations necessitated by complications such as pulmonary issues, growth failure, and orthopedic problems, and common supportive therapies such as ventilators, feeding, secretion suction, and orthosis support (10–13).

As of now, the FDA has approved three gene therapies for SMA treatment: Nusinersen (Spinraza®; Biogen), Onasemnogene abeparvovec (Zolengesma®; Novartis), and Risdiplam (Evrysdi®; F. Hoffmann-La Roche) (10). Despite their high cost, these therapies have demonstrated effectiveness by improving patient outcomes (14).

Despite ongoing research, the exact prevalence of SMA is difficult to ascertain. However, estimates suggest that it ranges from 1 to 2 per 100,000 individuals, with an incidence of 8–10 per 100,000 live births (15). Higher prevalence rates have been observed in Middle Eastern countries such as Saudi Arabia, where recent studies estimated roughly 2,265 SMA patients, possibly due to prevalent consanguineous marriages (16).

The varying severity of SMA has significant impacts on patients and their families, leading to financial strain, psychological challenges, sleep disturbances, and social limitations (17, 18). Comprehensive support mechanisms are needed, including psychological counseling, legal advice, genetic counseling, and family planning.

The economic burden of SMA is considerable and appears to be rising. The mean annual per-patient total cost of illness (direct medical costs, non-medical costs, and informal care) from the societal perspective varies between countries ranging from $97,300 (SMA type III) to $243,930 (SMA type I) in Australia, $60,770 (SMA type III) to $124,920 (SMA type I) in Germany, and $17,790 (SMA type III) to $39,520 (SMA type I) in Italy (19). Canadian research involving over 900 patients and caregivers highlighted significant costs to families of SMA patients, with median health expenditures for assistive devices, healthcare professional services, and accommodation and travel, along with a notable negative impact on patient quality of life (20). Although SMA is a rare health condition, its incidence rate is believed to be increasing in the kingdom mainly due to high rate of consanguinity and allocating more financial resources for preventative (e.g., premarital screening) and early detection and treatment measures (e.g., newborn screening, early initiation of therapy) is necessary (16).

Details on the specific characteristics of patients affected, the financial costs of the disease, the types of treatments provided, and the emotional state of caregivers, particularly in countries such as Saudi Arabia, are scarce (8). Future research should aim to evaluate and portray a more comprehensive landscape of the burden of illness across all SMA types. This endeavor will likely provide valuable insight for future healthcare planning and support for both SMA patients and their caregivers.

Moreover, there is a scarcity of information about how the disease impacts the population of Saudi Arabia specifically. This includes, but is not limited to, the individual characteristics of affected patients, the financial implications of the disease, the types of treatments administered, and the psychological state of caregivers.

Given the significance of these aspects, a detailed cost analysis is indispensable. This would enable a comparison between the expenditures associated with future potential curative therapies and the present palliative treatments, providing a clearer financial perspective on SMA management.

It would also be insightful to determine the rates of hospitalization and usage of mechanical ventilation. These metrics could serve as indicators of the disease's severity within the affected population. Furthermore, an assessment of the emotional toll on caregivers is needed to illuminate their perceptions, expectations, and strategies while caring for family members with SMA.

For the individuals afflicted with SMA, it is crucial to evaluate significant factors such as the health-related quality of life (HRQoL). This examination should encompass both the obstacles specific to the disease and the personal burdens it imposes.

At present, the development of innovative treatments for SMA management is progressing rapidly. It's therefore critical to conduct an in-depth study on how these advancements are influencing patients of SMA, particularly in specific cultural settings like Saudi Arabia where high rates consanguineous marriages are prevalent and some cultural beliefs of genetic risk factors.

With this in mind, we aim to provide a more comprehensive understanding of the burden of illness across all types of SMA.

The anticipated results could inform and enhance future healthcare planning, benefiting both patients and their caregivers.

2 Materials and methods

2.1 Study design

This study employed a cross-sectional design, leveraging questionnaires and proxy interviews to survey patients with SMA. The patient pool was selected using a snowball sampling technique from various caregivers attending the neurology clinics at King Khalid University Hospital in Riyadh, Saudi Arabia.

Inclusion in the study was predicated on the patients' confirmed diagnosis of SMA, as reported by their caregivers and verified by their treating neurologists. Exclusion criteria encompassed patients residing outside of Saudi Arabia and cases where caregivers were under the age of 18 years old. In this study, the caregivers of the patients served as proxy respondents, providing crucial data on behalf of the patients they care for.

2.2 Data collection

In this study, an interviewer-administered telephone survey was conducted, facilitated by three trained interviewers who engaged in data collection. An interview protocol was devised, and adherence to this protocol was ensured through role-playing exercises, aiming to minimize the risk of interviewer bias. Data collection commenced on July 26, 2022, and concluded on March 21, 2023.

The survey gathered sociodemographic characteristics of patients and caregivers, including factors such as age, gender, educational attainment, monthly income, geographical location, and number of siblings. Additionally, the family history of SMA in patients, the presence of SMA among siblings, the birth order of the patient within their family, and the educational level of the patient were recorded.

Furthermore, patient medical characteristics were documented. This information comprised age at diagnosis, the type of treatment received (supportive care only, Nusinersen, Onasemnogene abeparvovec-xioi, or Risdiplam), dependence on mechanical ventilation, instances of hospitalization in the past year, and participation in a regular physiotherapy program (21).

Out-of-pocket expenditures were assessed using a newly translated Arabic version of the 32-item CoPAQ. This translation adhered to the principles of good practice for the translation and cultural adaptation process for patient-reported outcome measures (22). The CoPAQ includes different questions that inquired about the patient's and caregiver's out-of-pocket expenditures related to the health condition of patient that were not covered by insurance or public assistance, such as, transportation, travels, parking fees, accommodation, prescription and non-prescription medications, dietary supplements, home care services (e.g., rehabilitation), medical devices, home renovation to accommodate patients' health condition, out-of-pocket expenses for healthcare services (e.g., copayment, coinsurance, deductible, and full cost payment for lab test or imaging studies, printing medical reports or certificates, dental services, osteopathy, etc.…), childcare, and pet care.

Patients' quality of life was evaluated using the Arabic version of the EuroQol (EQ-5D-5L) questionnaire among patients aged ≥12 years. Moreover, patients' self-rated health status was assessed using the visual analog scale of EuroQol (EQ-VAS) (23, 24). This version includes five domains: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression. Each domain has five possible levels, providing a comprehensive overview of the HRQoL among SMA patients. Caregivers served as proxy respondents for this assessment.

To gain insight into the mental health of the caregivers, their levels of depression and anxiety were evaluated using the Arabic versions of the Patient Health Questionnaire (PHQ-9) and General Anxiety Disorder-7 (GAD-7) tools, respectively (25, 26). The PHQ-9 consists of 9 items and is widely used in screening individuals on their levels of depression over the last 14 days, while the GAD-7 consists of seven items and screens individuals on their levels of anxiety over the last 14 days (25, 26). High scores in PHQ-9 and GAD-7 scales indicate higher levels of depression and anxiety, respectively.

Lastly, caregivers' health literacy was assessed using the Arabic version of the Single-Item Literacy Screener (SILS). According to this assessment, those who frequently, often, or sometimes required assistance with reading and understanding a medication leaflet were considered to have marginal literacy. Conversely, those who rarely or never needed such help were categorized as having adequate literacy (27, 28).

2.3 Data analysis

The requisite minimum sample size was calculated based on the difference in the EQ-VAS score for patients on FDA approved medications for SMA vs. their counterparts on supportive care using an alpha level (α) of 0.05, beta level (β) of 0.2, a large effect size as indicated by Cohen's d of 0.8, and a power of 80%. Based on these parameters, the minimum sample size was determined to be 52 SMA patients. Participant characteristics were analyzed using various descriptive statistical measures, including mean, median, standard deviation, interquartile range, as well as frequencies and percentages. Further inferential analysis was conducted using tests such as the Chi-Square test, Fisher's exact test, Student's t-test, and Analysis of Variance (ANOVA), as pertinent to the data set. The depression levels of the participants were categorized into five tiers, namely, minimal depression, mild depression, moderate depression, moderately severe depression, and severe depression, based on respective PHQ-9 scores of 5, 10, 15, and 20 (29). Anxiety levels were categorized into four tiers: minimal anxiety (GAD-7 scores 0–4), mild anxiety (GAD-7 scores 5–9), moderate anxiety (GAD-7 scores 10–14), and severe anxiety (GAD-7 scores 15 or above) (30). Only complete data were included in the analysis and no imputation was conducted. Cost in Saudi Riyals (SAR) was converted to United States Dollars (USD) using the fixed currency conversion rate of SAR 3.75 per USD 1. All statistical analyses were executed using the SAS® version 9.4 software suite (SAS Institute,

Cary, NC, USA). Graphical representations were generated using Microsoft® Excel 2016.

2.4 Ethical considerations

This study received formal approval from the Institutional Review Board (IRB) of King Saud University Medical City (Approval of Research Project No. E-22-6955), located in Riyadh, Saudi Arabia. Strict data access protocols were implemented to ensure the confidentiality and security of the patients' collected data, with access granted solely to the investigators associated with this study. All collected data were stored in a secure and safeguarded location. Personal identifiers, such as national ID numbers, were not collected, further upholding the anonymity of the participants. This research strictly adhered to the ethical principles outlined in the Helsinki Declaration, thereby ensuring the protection and respect of participant rights and welfare.

3 Results

3.1 Patients' sociodemographic characteristics

A total of 64 caregivers of SMA patients were identified and gave consent to participate in the study. The patients they cared for were divided into SMA types I ($n = 23$), II ($n = 19$), III ($n = 19$), and IV ($n = 3$). The mean patient age was ~13 years. However, patients with type I SMA were significantly younger than those with other types of SMA, with respective mean ages of 3.68 vs. 13.88 years, 22.52 years, and 17.67 years for types II, III, and IV, respectively (p-value < 0.0001).

Approximately 53% of the patients were males, and no significant differences were observed in gender distributions across the different types of SMA. All of the participants were Arab and a considerable majority of the patients were of Saudi nationality (93.75%), residing in the three most densely populated regions—Riyadh, Makkah, and the Eastern regions—which accounted for 82.82% of the patients. Furthermore, 85.94% of these patients resided in urban areas.

It was noted that patients with type I SMA had fewer siblings compared to their counterparts with other types of SMA. While the majority of patients did not report a family history of SMA (76.56%), ~48% did have siblings who were also diagnosed with the same type of SMA.

None of the patients with type I SMA were enrolled in formal schools since the majority (78.22%) were under 5 years of age. In contrast, patients with types II, III, and IV were engaged in different levels of formal education (p-value < 0.0001). These characteristics are summarized in Table 1.

3.2 Patients' medical characteristics

As expected, patients with SMA type I were diagnosed at a younger age compared to those with other types of SMA (1.29 vs. 3.29 years, 8.78 years, and 10.33 years for types II, III, and IV, respectively, p-value $= 0.0003$).

About 52% of patients were administered FDA-approved orphan medicinal products specific to SMA treatment, such as Nusinersen, Onasemnogene abeparvovec-xioi, and Risdiplam. Among these, the vast majority (90.91%) were treated with Nusinersen.

A significant proportion of type I patients (87%) were dependent on mechanical ventilation, contrasting with only 15.79% of patients with type II SMA, and none with types III and IV (p-value < 0.0001). Similarly, the majority of type I patients had been hospitalized during the previous 12 months, compared to 47.37, 26.32, and 0.0% for types II, III, and IV, respectively (p-value < 0.0001).

Most patients with types I, II, and IV were engaged in regular physical therapy programs, whereas a majority of type III patients were not (68.42%; p-value $= 0.0181$) as shown in Table 2. Interestingly, all type I SMA patients on supportive care were ventilator dependent, in comparison to 72.73% of those treated with FDA-approved orphan medical products for SMA. However, this difference was not statistically significant (p-value $= 0.0932$). Similarly, there was no noticeable difference in the percentages of ventilator-dependent patients among type II SMA patients based on the type of treatment (supportive care only vs. FDA-approved drugs for SMA; p-value $= 1.0000$). This is graphically represented in Figure 1.

There were no significant differences in the rates of hospitalization in the past 12 months among SMA type I patients on supportive care only or those on FDA-approved drugs for SMA (91.67 vs. 90.91%, p-value $= 0.9486$). Even though the rates of hospitalization in the past 12 months among patients with types II and III SMA treated with FDA-approved drugs for SMA were higher than their counterparts on supportive care only (53.85 vs. 33.33% and 33.33 vs. 20.00%, respectively), this difference did not reach statistical significance (p-value $= 0.6285$).

3.3 Proxy-reported health-related quality of life

The number of patients aged ≥12 years who answered the EQ-5D-5L questions on the five domains were 22 patients. All patients with SMA types I and II, according to their EQ-5D-5L responses, were incapable of walking, contrasting sharply with only 50.0% of type III patients and none of the type IV patients. Similarly, all of the patients with type I and II were unable to take care of themselves and the majority were unable to perform their usual activities as shown in Table 3.

Utilizing the EQ-VAS, which assessed overall health on a scale from 0 (worst imaginable health) to 100 (best imaginable health) for 52 SMA patients from different age groups (type I = 16, type II = 17, type III = 16, type IV = 3), the mean scores were 33.85 for type I, 38.57 for type II, 52.04 for type III, and 40.66 for type IV patients (p-value $= 0.0137$). Interestingly, when EQ-VAS scores were stratified across SMA types and treatment (supportive care only vs. treatment

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TABLE 1 Patient baseline characteristics.

Characteristic	Type I (*N* = 23)	Type II (*N* = 19)	Type III (*N* = 19)	Type IV (*N* = 3)	*p*-value	Total (*N* = 64)
Age, *N* (%)						
<1 yr.	5 (21.74)	1 (5.26)	0 (0.0)	0 (0.0)	0.0001*	6 (9.38)
≥1–<2 yrs.	4 (17.39)	0 (0.0)	0 (0.0)	0 (0.0)		4 (6.25)
≥2–<5 yrs.	9 (39.13)	2 (10.53)	2 (10.53)	0 (0.0)		13 (20.31)
≥5–<10 yrs.	4 (17.39)	6 (31.58)	4 (21.05)	1 (33.33)		15 (23.44)
≥10–<18 yrs.	0 (0.0)	5 (26.32)	2 (10.53)	0 (0.0)		7 (10.94)
≥18 yrs.	1 (4.35)	5 (26.32)	11 (57.89)	2 (66.67)		19 (29.69)
Gender, *N* (%)						
Male	11 (47.83)	10 (52.63)	11 (57.89)	2 (66.67)	0.9244	34 (53.13)
Female	12 (52.17)	9 (47.37)	8 (42.11)	1 (33.33)		30 (46.88)
Region, *N* (%)						
Riyadh	5 (21.74)	9 (47.37)	4 (21.05)	1 (33.33)	0.4423	19 (29.69)
Makkah	5 (21.74)	5 (26.32)	6 (31.58)	2 (66.67)		18 (28.13)
Eastern region	9 (39.13)	4 (21.05)	3 (15.79)	0 (0.0)		16 (25.00)
Al-Jawf	1 (4.35)	0 (0.0)	0 (0.0)	0 (0.0)		1 (1.56)
Jazan	0 (0.0)	1 (5.26)	2 (10.53)	0 (0.0)		3 (4.69)
Al Qassim	0 (0.0)	0 (0.0)	1 (5.26)	0 (0.0)		1 (1.56)
Aseer	0 (0.0)	0 (0.0)	1 (5.26)	0 (0.0)		1 (1.56)
Tabuk	2 (8.70)	0 (0.0)	2 (10.53)	0 (0.0)		4 (6.25)
Hail	1 (4.35)	0 (0.0)	0 (0.0)	0 (0.0)		1 (1.56)
Nationality, *N* (%)						
Saudi	20 (86.96)	19 (100.0)	18 (94.74)	3 (100.0)	0.6259	60 (93.75)
Non-Saudi	3 (13.05)	0 (0.0)	1 (5.26)	0 (0.0)		4 (6.25)
Number of siblings, *N* (%)						
None	5 (21.74)	0 (0.0)	1 (5.26)	0 (0.0)	0.3251	6 (9.38)
1-2	3 (13.04)	2 (10.53)	2 (10.53)	0 (0.0)		7 (10.94)
3-4	12 (52.17)	9 (47.37)	12 (63.16)	2 (66.67)		35 (54.69)
>4	3 (13.04)	8 (42.11)	4 (21.05)	1 (33.33)		16 (25.00)
Living in urban or rural areas, *N* (%)						
Urban	18 (78.26)	19 (100.00)	15 (78.95)	3 (100.00)	0.1417	55 (85.94)
Rural	5 (21.74)	0 (0.0)	4 (21.05)	0 (0.0)		9 (14.06)
Family history of SMA, *N* (%)						
Yes	4 (17.39)	5 (26.32)	5 (26.32)	1 (33.33)	0.9396	15 (23.44)
No	19 (82.61)	14 (73.68)	14 (73.68)	2 (66.66)		49 (76.56)
Have siblings with SMA, N (%)						
Yes	9 (39.13)	12 (63.16)	7 (36.84)	3 (100.0)	0.0906	31 (48.44)
No	14 (60.87)	7 (36.84)	12 (63.16)	0 (0.0)		33 (51.56)
Which type of SMA your sibling was diagnosed with?						
SMA type I	6 (26.09)	0 (0.0)	0 (0.0)	0 (0.0)	<0.0001*	6 (9.38)

(Continued)

TABLE 1 (Continued)

Characteristic	Type I ($N = 23$)	Type II ($N = 19$)	Type III ($N = 19$)	Type IV ($N = 3$)	*p*-value	Total ($N = 64$)
SMA type II	1 (4.35)	9 (47.37)	0 (0.0)	2 (66.66)		12 (18.75)
SMA type III	0 (0.0)	0 (0.0)	6 (31.58)	0 (0.0)		6 (9.38)
SMA type IV	0 (0.0)	0 (0.0)	1 (5.26)	1 (33.33)		2 (3.13)
Patient order among his/her siblings, *N* (%)	2.65 ± 1.55	3.53 ± 2.65	3.05 ± 1.72	4.33 ± 0.58	0.3626	3.11 ± 1.98
First	8 (34.78)	4 (21.05)	5 (26.32)	0 (0.0)	0.1364	17 (26.56)
Second or third	7 (30.43)	8 (42.11)	7 (36.84)	0 (0.0)		22 (34.38)
Fourth or fifth	8 (34.78)	3 (15.79)	6 (31.58)	3 (100.0)		20 (31.25)
Sixth or more	0 (0.0)	4 (21.05)	1 (5.26)	0 (0.0)		5 (7.81)
Patient current educational level						
Not in school	23 (100.00)	4 (21.05)	3 (11.11)	0 (0.00)	<0.0001*	28 (43.75)
Kindergarten	0 (0.00)	0 (0.00)	1 (5.26)	0 (0.00)		2 (3.13)
Elementary	0 (0.00)	10 (52.63)	4 (21.05)	1 (33.33)		15 (23.44)
Intermediate	0 (0.00)	1 (5.26)	1 (5.26)	0 (0.00)		3 (4.69)
Secondary	0 (0.00)	2 (10.53)	4 (21.05)	1 (33.33)		7 (10.94)
College	0 (0.0)	2 (10.53)	6 (31.58)	1 (33.33)		9 (14.06)

*p-value < 0.05.

TABLE 2 Patient medical characteristics.

Characteristic	Type I ($N = 23$)	Type II ($N = 19$)	Type III ($N = 19$)	Type IV ($N = 3$)	*p*-value	Total ($N = 64$)
Age at diagnosis (yrs.), mean ± SD	1.29 ± 2.16	3.29 ± 3.89	8.78 ± 7.94	10.33 ± 9.01	0.0003*	4.54 ± 6.16
Type of treatment received, *N* (%)						
Only supportive care	12 (52.17)	6 (31.58)	10 (52.63)	3 (100.0)	0.3243	31 (48.44)
Nusinersen	9 (39.13)	12 (63.16)	9 (47.37)	0 (0.0)		30 (46.88)
Onasemnogene abeparvovec-xioi	1 (4.35)	1 (5.26)	0 (0.0)	0 (0.0)		2 (3.13)
Risdiplam	1 (4.35)	0 (0.0)	0 (0.0)	0 (0.0)		1 (1.56)
Patient is dependent on mechanical ventilation, *N* (%)						
No	3 (13.04)	16 (84.21)	19 (100.0)	3 (100.0)	<0.0001*	41 (64.06)
Yes	20 (86.96)	3 (15.79)	0 (0.0)	0 (0.0)		23 (35.94)
Rates of hospitalization in the past 12 months, *N* (%)						
Yes	21 (91.30)	9 (47.37)	5 (26.32)	0 (0.0)	<0.0001*	35 (54.69)
No	2 (8.70)	10 (52.63)	14 (73.68)	3 (100.0)		29 (45.31)
Rates of intensive care unit (ICU) hospitalization in the past 12 months, *N* (%)						
Yes	19 (82.61)	9 (47.37)	5 (26.32)	0 (0.0)	0.0004*	33 (51.56)
No	4 (17.39)	10 (52.63	14 (73.68)	3 (100.0)		31 (48.44)
Patient is enrolled in regular physical therapy? *N* (%)						
Yes	15 (65.22)	15 (78.95)	6 (31.58)	2 (66.67)	0.0181*	38 (59.38)
No	8 (34.78)	4 (21.05)	13 (68.42)	1 (33.33)		26 (40.63)

*p-value < 0.05.

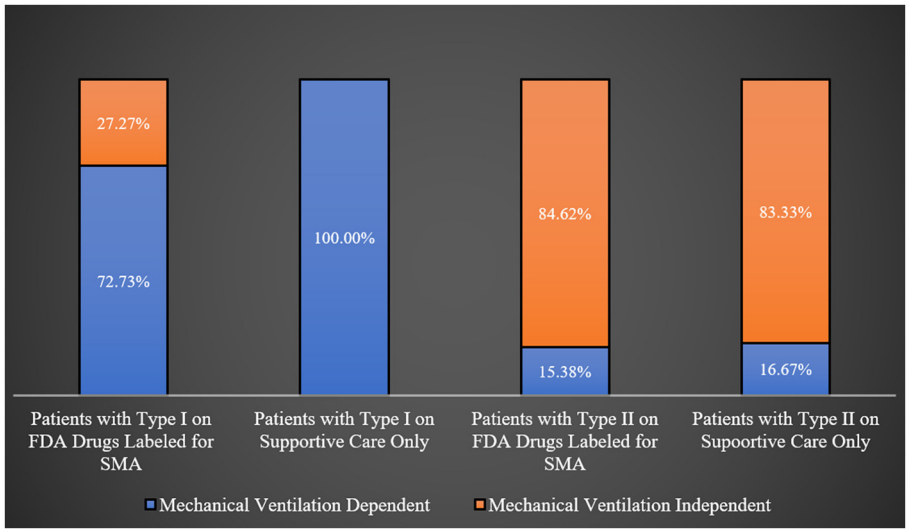

FIGURE 1
Mechanical ventilation status across patients on different treatment regimens.

with FDA-approved drugs for SMA), the higher mean score observed in type III patients compared to other types of SMA was no longer apparent. However, type III patients managed solely with supportive care such as nutritional and respiratory support registered a mean EQ-VAS score of 36.75 ± 11.40, compared to 67.33 ± 14.15 among those treated with FDA-approved drugs for SMA (p-value < 0.0001) as demonstrated in Figure 2.

3.4 Caregivers' baseline characteristics and financial burden

In this study, the majority of caregivers for SMA patients were found to be parents (82.18%), married (90.63%), between the ages of 20 and 30 (75.01%), holding an associate degree or higher (51.56%), and possessing adequate health literacy (65.63%). Interestingly, 53.13% of the caregivers did not have a paid job, and most were earning <$1,600 a month (i.e., this includes the main caregiver's income and does not include household income) which is deemed below the average national monthly income (e.g., USD 2,730). A significant majority (85.94%) did not receive any formal education or training on how to provide care for SMA patients, and 59.38% traveled to seek medical consultation for their patient(s) with SMA. Even so, 93.75% did not experience income loss (for example, from missing work days due to their patient's illness), as highlighted in Table 4. Nevertheless, more than half of the caregivers reported experiencing some form of financial stress.

The caregivers for SMA type I patients, followed by those for type II patients, had significantly higher mean PHQ-9 scores than caregivers for other types of SMA patients (p-value = 0.0155). This is visibly demonstrated in Figure 3, showing 34.78 and 31.58% of caregivers for SMA type I and II patients, respectively, experiencing moderate to moderately severe depression, compared to just 15.79% of caregivers for type III patients.

However, no significant difference was detected in the mean GAD-7 scores among caregivers for patients with different types of SMA, nor in levels of anxiety, as illustrated in Figure 4.

Interestingly, caregivers for SMA type III patients reported the highest mean out-of-pocket expenditures. The mean total out-of-pocket expenditures per patient per year amounted to USD 7,099.77 for type I, USD 3,395.75 for type II, USD 19,055.58 for type III, and USD 444.45 for type IV. These expenses encompassed both medical costs (for instance, private clinic visits, over-the-counter medications, and medical devices) and non-medical costs (such as home renovations or purchasing a specially-equipped vehicle).

4 Discussion

To the best of our knowledge, this study represents the first attempt to investigate the socioeconomic burden experienced by both SMA patients and their caregivers in Saudi Arabia. By delving into the various aspects of the socioeconomic impact of SMA, we aimed to contribute valuable insights to the existing body of knowledge in this field.

Although a previously published study examined the impact of Nusinersen treatment on the HRQoL of SMA patients in Saudi Arabia, using caregivers as proxy respondents, raised concerns regarding the effectiveness of Nusinersen in improving HRQoL, this study did not assess the psychological impact of SMA on caregivers or considered the financial burden associated with the illness. Consequently, our research comprehensively evaluated the psychological wellbeing of caregivers and quantify the out-of-pocket expenditures related to SMA management (31).

Moreover, the economic burden of SMA and the cost-effectiveness of treatments have been evaluated in different countries. These studies provide insights into the direct medical

TABLE 3 Proxy-reported EuroQol-5-D-5-L scores for those aged 12 years and above.

Characteristic	Type I ($N = 1$)	Type II ($N = 9$)	Type III ($N = 10$)	Type IV ($N = 2$)	*p*-value	Total ($N = 22$)
EQ-5D-5L domains						
Mobility, *N* (%)						
I have no problems in walking about	0 (0.0)	0 (0.0)	1 (10.0)	2 (100.0)	0.6015	3 (13.64)
I have slight problems in walking about	0 (0.0)	0 (0.0)	2 (20.0)	0 (0.0)		2 (9.09)
I have moderate problems in walking about	0 (0.0)	0 (0.0)	1 (10.0)	0 (0.0)		1 (4.55)
I have severe problems in walking about	0 (0.0)	0 (0.0)	1 (10.0)	0 (0.0)		1 (4.55)
I am unable to walk about	1 (100.0)	9 (100.0)	5 (50)	0 (0.0)		15 (68.18)
Self-care, *N* (%)						
I have no problems washing or dressing myself	0 (0.0)	0 (0.0)	2 (20.00)	0 (0.0)	0.4299	2 (9.09)
I have slight problems washing or dressing myself	0 (0.0)	0 (0.0)	2 (20.00)	0 (0.0)		2 (9.09)
I have moderate problems washing or dressing myself	0 (0.0)	0 (0.0)	2 (20.00)	2 (100.0)		4 (18.18)
I have severe problems washing or dressing myself	0 (0.0)	0 (0.0)	1 (10.00)	0 (0.0)		2 (9.09)
I am unable to wash or dress myself	1 (100.0)	9 (100.0)	3 (30.00)	0 (0.0)		13 (59.09)
Usual activities (e.g., work, study, housework, family, or leisure activities), *N* (%)						
I have no problems doing my usual activities	0 (0.0)	0 (0.0)	1 (10.00)	0 (0.0)	0.0425	1 (4.55)
I have slight problems doing my usual activities	0 (0.0)	0 (0.0)	3 (30.00)	2 (100.0)		5 (22.73)
I have moderate problems doing my usual activities	0 (0.0)	0 (0.0)	2 (20.00)	0 (0.0)		2 (9.09)
I have severe problems doing my usual activities	0 (0.0)	2 (22.22)	2 (20.00)	0 (0.0)		4 (18.18)
I am unable to do my usual activities	1 (100.0)	7 (77.78)	2 (20.00)	0 (0.0)		10 (45.45)
Pain/discomfort, *N* (%)						
I have no pain or discomfort	0 (0.0)	1 (11.11)	4 (40.00)	0 (0.0)	0.1718	5 (22.73)
I have slight pain or discomfort	1 (100.0)	4 (44.44)	1 (10.00)	2 (100.0)		8 (36.36)
I have moderate pain or discomfort	0 (0.0)	4 (44.44)	2 (20.00)	0 (0.0)		6 (26.92)
I have severe pain or discomfort	0 (0.0)	0 (0.0)	1 (10.00)	0 (0.0)		1 (4.55)

(Continued)

TABLE 3 (Continued)

Characteristic	Type I (*N* = 1)	Type II (*N* = 9)	Type III (*N* = 10)	Type IV (*N* = 2)	*p*-value	Total (*N* = 22)
I have extreme pain or discomfort	0 (0.0)	0 (0.0)	2 (20.00)	0 (0.0)		2 (9.09)
Anxiety/depression, *N* (%)						
I am not anxious or depressed	1 (100.00)	5 (55.56)	6 (60.00)	0 (0.0)	0.5237	12 (54.55)
I am slightly anxious or depressed	0 (0.0)	2 (22.22)	1 (10.00)	2 (100.0)		5 (22.73)
I am moderate anxious or depressed	0 (0.0)	2 (22.22)	1 (10.00)	0 (0.0)		3 (13.64)
I am severely anxious or depressed	0 (0.0)	0 (0.0)	1 (10.00)	0 (0.0)		1 (4.55)
I am extremely anxious or depressed	0 (0.0)	0 (0.0)	1 (10.00)	0 (0.0)		1 (4.55)

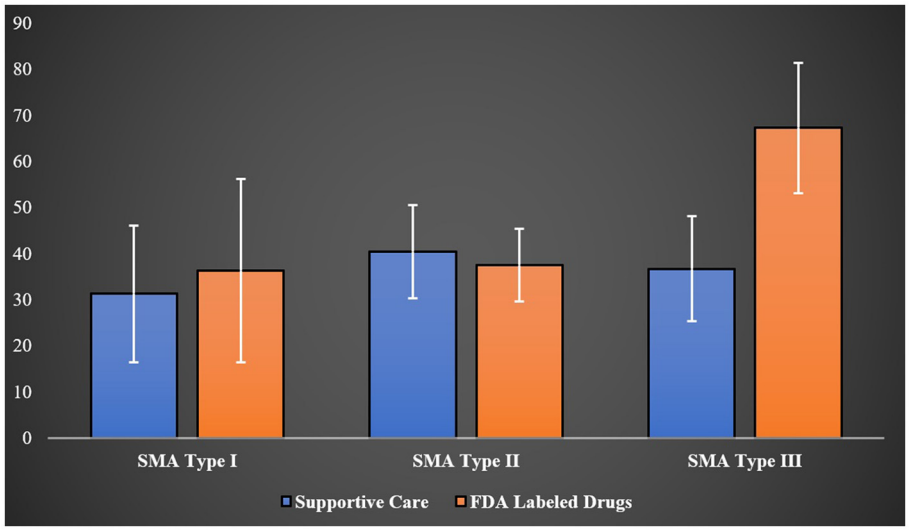

FIGURE 2
The EQ-VAS scores for patients with type I, II, and III of SMA.

and non-medical costs associated with SMA, as well as the impact on patients and caregivers.

A systematic review conducted in 2020 identified a range of cost-effectiveness evaluations for SMA treatments. The incremental cost-effectiveness ratio (ICER) for nusinersen, one of the treatments assessed, varied from $210,095 to $1,150,455 per quality-adjusted life years (QALY) gained (14). Furthermore, another study found that the mean per-patient annual direct medical costs ranged from $3,320 to $324,410, depending on the type of SMA and the country (19).

In terms of the burden on patients and caregivers, a study conducted in Canada highlighted the impaired quality of life experienced by SMA patients. Caregivers reported the need for various forms of support, such as assistive devices and health professional services. They also faced challenges in terms of personal plans, sleep disturbances, and work adjustments (20). Similarly, a study in Hong Kong revealed a high healthcare burden and cumulative life costs for SMA patients, particularly those with type 1 and type 2 SMA, who did not receive disease-modifying treatment (32).

In our own research conducted in Saudi Arabia, we observed specific patterns within different SMA types. Type 1 patients had higher rates of sibling concordance, ICU hospitalization, and a greater need for mechanical support. Conversely, Type 3 patients exhibited a better quality of life. Type 1 patients' caregivers also showed higher depression scores, while caregivers

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TABLE 4 Caregivers' characteristics and SMA financial burden.

Characteristic	Type I ($N = 23$)	Type II ($N = 19$)	Type III ($N = 19$)	Type IV ($N = 3$)	*p*-value	Total ($N = 64$)
Relationship of main caregiver to patient, *N* (%)						
Mother	13 (56.52)	10 (52.63)	12 (63.16)	1 (33.33)	0.0522	36 (56.25)
Father	10 (43.48)	4 (21.05)	2 (10.53)	1 (33.33)		17 (26.56)
Sibling	0 (0.0)	2 (10.53)	2 (10.53)	0 (0.0)		4 (6.25)
Spouse	0 (0.0)	1 (5.26)	2 (10.53)	1 (33.33)		4 (6.25)
Maid[†]	0 (0.0)	2 (10.53)	1 (5.26)	0 (0.0)		3 (4.69)
Marital status of the main caregiver, *N* (%)						
Single	0 (0.00)	1 (5.26)	1 (5.26)	0 (0.00)	0.0524	2 (3.13)
Married	23 (100.0)	15 (78.95)	17 (89.47)	3 (100.0)		58 (90.63)
Divorced	0 (0.00)	3 (15.79)	1 (5.26)	0 (0.00)		4 (6.25)
Age of the main caregiver, *N* (%)						
<20 yrs.	2 (8.69)	0 (0.0)	0 (0.0)	0 (0.0)	0.1618	2 (3.13)
20–30 yrs.	6 (26.09)	2 (10.53)	2 (10.53)	0 (0.0)		10 (15.63)
31–40 yrs.	14 (60.87)	12 (63.16)	9 (47.37)	3 (100.0)		38 (59.38)
41–50 yrs.	1 (4.35)	3 (15.79)	4 (21.05)	0 (0.0)		8 (12.5)
51–60 yrs.	0 (0.0)	2 (10.53)	3 (15.79)	0 (0.0)		5 (7.81)
61–70 yrs.	0 (0.0)	0 (0.0)	1 (5.26)	0 (0.0)		1 (1.56)
Educational level of the caregiver, *N* (%)						
No formal education	0 (0.0)	0 (0.0)	1 (5.26)	0 (0.0)	0.4367	1 (1.56)
Elementary school	3 (13.04)	0 (0.0)	2 (10.53)	0 (0.0)		5 (7.81)
Intermediate school	0 (0.0)	1 (5.26)	0 (0.0)	0 (0.0)		1 (1.56)
High school diploma	10 (43.48)	5 (26.32)	9 (47.37)	0 (0.0)		24 (37.5)
Associate degree	2 (8.69)	1 (5.26)	1 (5.26)	0 (0.0)		4 (6.25)
College degree	6 (26.09)	11 (57.89)	6 (31.58)	3 (100.0)		26 (40.63)
Postgraduate degree	2 (8.69)	1 (5.26)	0 (0.0)	0 (0.0)		3 (4.69)
Health literacy, *N* (%)						
Adequate	12 (52.17)	16 (84.21)	12 (63.16)	2 (66.67)	0.1642	42 (65.63)
Marginal	11 (47.83)	3 (15.79)	7 (36.84)	1 (33.33)		22 (34.38)
Do you have a paying job?						
Yes	11 (47.83)	11 (57.89)	7 (36.84)	1 (33.33)	0.4973	30 (46.88)
No	12 (52.17)	8 (42.11)	12 (63.16)	2 (66.66)		34 (53.13)
Monthly income (USD), *N* (%)						
$0–$800	12 (52.17)	8 (42.11)	9 (47.37)	3 (100.0)	0.5497	32 (50.0)
$800–$1,600	5 (21.74)	3 (15.79)	3 (15.79)	0 (0.0)		11 (17.19)
$1,600–$2,666.67	3 (13.04)	1 (5.26)	3 (15.79)	0 (0.0)		7 (10.94)
$2,666.67–$4,000	1 (4.35)	3 (15.79)	2 (10.53)	0 (0.0)		6 (9.38)
$4,000–$5,333.33	1 (4.35)	3 (15.79)	1 (5.26)	0 (0.0)		5 (7.81)
>$5,333.33	1 (4.35)	1 (5.26)	0 (0.0)	0 (0.0)		2 (3.125)
Did you receive education or training sessions on how to care for SMA patients? *N* (%)						
Yes	6 (26.09)	1 (5.26)	2 (10.53)	0 (0.0)	0.2434	9 (14.1)

(Continued)

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TABLE 4 (Continued)

Characteristic	Type I ($N = 23$)	Type II ($N = 19$)	Type III ($N = 19$)	Type IV ($N = 3$)	*p*-value	Total ($N = 64$)
No	17 (73.91)	18 (94.74)	17 (89.47)	3 (100.0)		55 (85.94)
Did you travel to seek medical consultation for your relative? *N* (%)						
Yes	13 (56.52)	10 (52.63)	14 (73.68)	1 (33.33)	0.2038	38 (59.38)
No	10 (43.48)	9 (47.37)	5 (26.32)	2 (66.66)		26 (40.63)
Did you suffer any income loss due to your relative's disease? *N* (%)						
Yes	2 (8.70)	2 (10.53)	0 (0.0)	0 (0.0)	0.7389	4 (6.25)
No	21 (91.30)	17 (89.47)	19 (100.0)	3 (100.0)		60 (93.75)
What was the reason behind the income loss? *N* (%)						
Missed days or showing up late to work	2 (8.70)	1 (5.26)	0 (0.0)	0 (0.0)	1.00	3 (4.69)
Health insurance refusal to pay	0 (0.0)	1 (5.26)	0 (0.0)	0 (0.0)		1 (1.56)
Are you under financial strain? *N* (%)						
Refrain from answering	5 (21.74)	4 (21.05)	8 (42.11)	1 (33.33)	0.0345	18 (28.13)
Not at all	4 (17.39)	2 (10.53)	6 (31.58)	0 (0.0)		12 (18.75)
Sometimes	6 (26.09)	3 (15.79)	0 (0.0)	2 (66.66)		11 (17.19)
Often	3 (13.04)	5 (26.32)	0 (0.0)	0 (0.0)		8 (12.5)
Always	5 (21.74)	5 (26.32)	5 (26.32)	0 (0.0)		15 (23.44)
Patient Health Questionnaire-9 (PHQ-9), Mean ± SD	9.88 ± 4.77	6.65 ± 4.95	6.38 ± 3.92	3.00 ± 3.58	0.0155*	7.56 ± 4.79
Generalized Anxiety Disorder-7 (GAD-7), Mean ± SD	6.79 ± 5.45	4.18 ± 4.30	5.38 ± 4.72	6.0 0 ± 4.97	0.3803	5.56 ± 4.81
Estimated total out of pocket expenditures since the time of diagnosis, mean ± SD (USD)	3,722.32 ± 6,087.38	13,492.49 ± 15,799.45	33,366.32 ± 73,775.52	3,111.11 ± 2,694.30	0.1399	15,394.75 ± 42,367.53

†Maids who are part of the household served as proxy respondents to the biological main caregivers (e.g., father, mother, sister, and brother) due to the difficulty of reaching the biological relatives most of the time. *p-value < 0.05.

of Type 3 patients reported higher out-of-pocket expenditures. The higher out-of-pocket expenditures reported by caregivers for patients with type III SMA is expected due to the higher survival rates compared to patients with types I and II and the earlier onset of the disease compared to patients with type IV (6–8). Moreover, it is worth noting that 48% of patients received supportive care, while others received SMA therapies. The overall findings underscored the significant socioeconomic burden imposed by SMA on patients and caregivers, emphasizing the need for increased attention from the healthcare system. Access to innovative therapies varied across different SMA types. Therefore, further efforts should be directed toward implementing screening programs and providing timely access to innovative therapies to mitigate the impact of SMA on individuals and society. These findings underscore the importance of providing patients with SMA with supplemental health insurance to cover other expenses that are mostly uncovered by the public healthcare coverage such as durable medical equipment. Additionally, there are other non-medical expenses, such as, vehicle modifications to accommodate needs of patients with disabilities and home remodeling for people's disabilities. Therefore, patients with rare and burdensome illnesses, such as SMA, should receive supplemental government financial support.

On the other hand, significant variation in the mean age across different subtypes of SMA was observed. Notably, the mean age for type III was observed to be higher in comparison to type I (22.52 ± 15.97 vs. 3.68 ± 5.16). These results align with previous studies that have consistently reported a mean age of onset of 5 months for

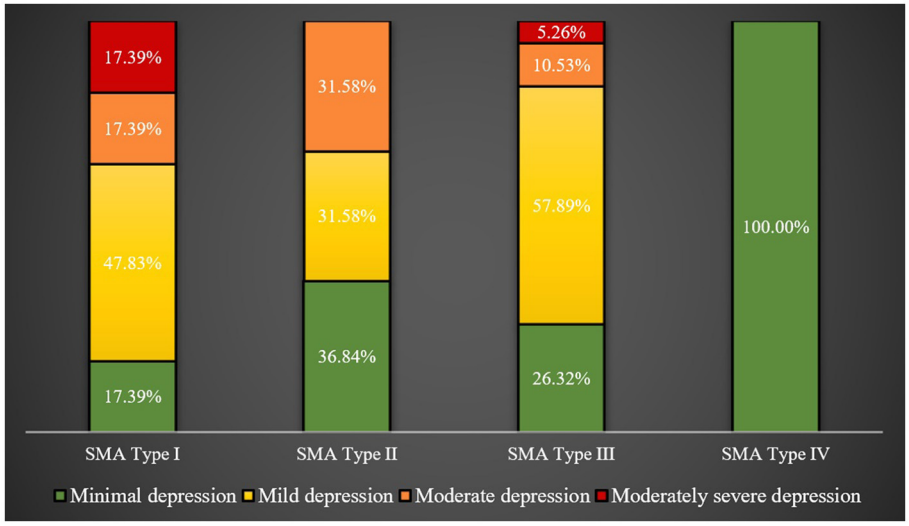

FIGURE 3
Rates of depression across caregivers of patients with different SMA types.

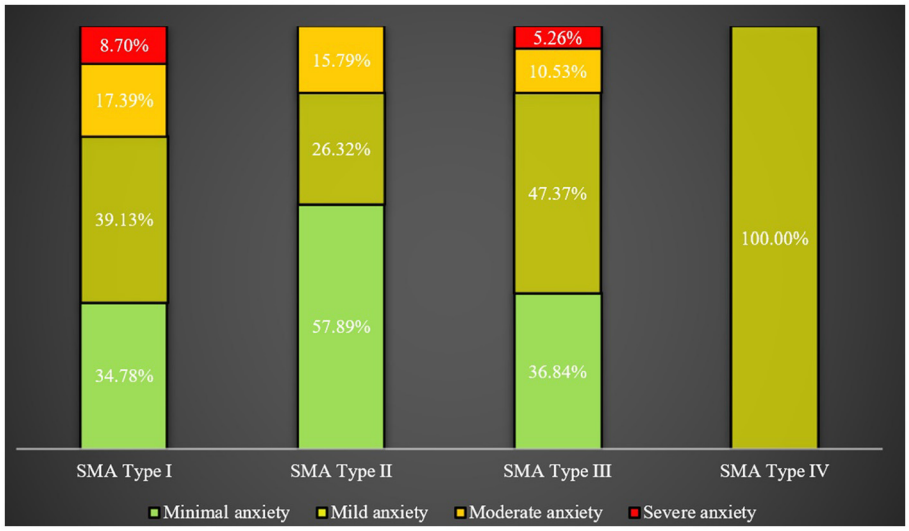

FIGURE 4
Rates of anxiety across caregivers of patients with different SMA types.

SMA type 1 patients, 11.5 months for type 2, 4.5 years for type 3, and 18 years for type 4 (33, 34).

Furthermore, it is worth noting that the mean age for type IV was found to be 17.67 ± 8.50. This discrepancy may be attributed to the relatively small number of patients with type IV SMA in our study sample, as there were only three participants compared to the other SMA types that had nearly 20 participants. Alternatively, this variation could potentially reflect the natural diversity present within the Saudi population. However, it is important to emphasize that a larger sample size in prospective studies would be needed to obtain more conclusive insights into this matter.

The age distribution corresponded with the age of diagnosis and educational level. Among SMA type I patients, the majority were children who had not yet started school. Similarly, in type II, most individuals were either in elementary school or had completed it. Interestingly, type III exhibited a wide distribution, with some patients as early as kindergarten and others already in college. However, this distribution does not necessarily align with age and may simply reflect the challenges faced by individuals with SMA in pursuing education.

Furthermore, this study revealed a notable trend in the Saudi population, where siblings tended to share the same subtype of SMA, as expected in a disease with a familial distribution. Similar findings have been reported in other studies. For instance, a cohort study examining 303 siblings identified between 1996 and 2016 reported that 84.8% of them exhibited subtype concordance. The

distribution of concordant subtypes in this study was as follows: Type I, 54.5%; Type II, 31.9%; Type III, 13.2%; Type IV, 0.4% (35). These findings provide further evidence of the familial nature of SMA and its impact on affected siblings.

SMA type 1 patients constituted the majority of individuals requiring mechanical ventilation (86.96% vs. 3% vs. 0% vs. 0%) due to the severe nature of the disease and its early onset at around 6 months of age. These patients typically experience a rapid disease course, leading to lifelong reliance on ventilatory support before the age of 2 (36). Consequently, it is not surprising that a significant proportion of SMA type 1 patients had a history of hospitalization (91.3% vs. 47.37% vs. 26.32% vs. 0%). Among the 21 SMA type 1 patients requiring mechanical ventilation, 19 of them were admitted to the intensive care unit (ICU). It is worth noting that current treatment guidelines for SMA, particularly types 1–3, emphasize the importance of early assessment of lung function and the implementation of supportive respiratory therapies (37). These include techniques such as air stacking, physiotherapy, mechanical insufflation, and mechanical exsufflation procedures. While patients with other subtypes may not have utilized mechanical ventilatory support, they may have employed less invasive respiratory devices like CPAP or oxygen tanks. However, this specific information was not collected from the study participants.

The majority of individuals across all SMA subtypes engaged in physiotherapy, regardless of disease severity, which is crucial for maximizing physical functionality. Physiotherapy helps improve posture, prevent joint immobility, and reduce muscle atrophy and weakness (38, 39). Guidelines recommend that all SMA patients have access to specialized neuromuscular centers, where they can receive regular evaluations and physiotherapy recommendations from skilled and experienced professionals every 6 months (40, 41). However, it is concerning that ∼41% of the study sample were not enrolled in regular physical therapy, as this may lead to worse clinical outcomes and a faster rate of disease progression, even among those receiving medications with FDA-labeled indications for SMA (42).

The availability of specialized physical therapy centers catering to SMA patients in Saudi Arabia is insufficient, and it is imperative for the Saudi Physical Therapy Association (SPTA) to prioritize this specific patient subset while promoting awareness about the prevalence of the disease within the country.

According to the proxy-reported EuroQol assessment, the HRQoL demonstrated improvement with increasing SMA subtype, with Type I and Type II patients reporting the lowest HRQoL, while Type III and Type IV patients showed relatively better HRQoL based on the EQ-5D-5-L and EQ-VAS scales.

In terms of mobility and self-care, all participants with Type I and Type II SMA were unable to walk or maintain self-care. Additionally, all Type I individuals reported an inability to perform usual daily activities, compared to 76.47% of Type II patients. On the other hand, ∼50% of Type III patients had either no problem, slight problem, or moderate problem in walking. These findings are consistent across the four other domains of the EQ-5D-5L questionnaire, except for anxiety/depression. Similar studies have shown better HRQoL among patients with Type III SMA compared to their counterparts with Type I and Type II SMA (43). Pain symptoms were not widespread, with only 43.75% of Type I patients reporting severe pain. While there was no significant association between anxiety/depression and SMA subtype or symptom severity, overall EQ-VAS values were higher for Type III and Type IV patients. Interestingly, another study conducted in Iran found no significant difference in HRQoL between Type II and Type III SMA patients (44).

The psychological wellbeing of caregivers showed that the majority experienced minimal to mild levels of anxiety and depression, regardless of the type of patient they were caring for. However, caregivers for patients with type I SMA exhibited higher PHQ-9 scores, which is intriguing considering that previous studies examining the psychological impact on caregivers of SMA patients often reported elevated levels of depression and anxiety (45, 46). This observation may be attributed to the cultural concept of filial piety, as Saudi Arabian culture and religion place significant emphasis on caring for the sick and older adults, regarding it as a noble and rewarding act (47).

Analysis of out-of-pocket expenditures incurred by SMA caregivers since the time of diagnosis revealed higher costs for type III, followed by type II, compared to type I. This is primarily due to the higher survival rates associated with type III and II SMA, in contrast, to type I, which typically has a life expectancy of <2 years (48). Additionally, it is worth noting that a significant portion of the out-of-pocket expenditures were not directly medically related. These expenses included purchasing care for individuals with special needs and making home renovations to accommodate the needs of the patients. This pattern may be influenced by the presence of universal healthcare coverage for citizens in Saudi Arabia, resulting in fewer medically-related out-of-pocket costs (49).

Regarding treatment options, all three therapies approved by the United States Food and Drug Administration (Nusinersen, Onasemnogene abeparvovec-xioi, and Risdiplam) have also been approved by the Saudi Food and Drug Authority (SFDA). However, the access to these therapies varied among the study participants, with Nusinersen being the most commonly utilized SMA therapy, accounting for ∼47% of cases. This discrepancy in utilization rates may be attributed to the fact that Nusinersen was the first SMA therapy approved by the USFDA in December 2016, while Onasemnogene abeparvovec-xioi received approval in May 2019, and Risdiplam in August 2020 (50).

Moreover, these findings shed light on the psychological wellbeing of caregivers, the financial burden they face, and the availability of SMA therapies in Saudi Arabia. Understanding these aspects is crucial for developing support systems and interventions that cater to the specific needs of caregivers and patients with SMA in the country.

Interestingly, no significant difference was observed in the rates of patients on mechanical ventilation or the rate of hospitalization in the past 12 months between patients managed with FDA-labeled therapies and those managed with supportive care alone. This finding aligns with a previously published study conducted in Saudi Arabia that reported similar results (31).

However, among SMA patients with type III, those managed with Nusinersen showed a significantly higher mean EQ-VAS score, which measures overall health-related quality of life (HRQoL), compared to those managed with supportive care alone. Conversely, no significant difference was found between

patients with type I and II managed with FDA-labeled therapies or supportive care. These findings are intriguing, as studies assessing HRQoL among patients managed with Nusinersen have not consistently demonstrated significant improvements across different SMA types, despite modest improvements observed in type I and II cases (31, 51, 52). However, these findings align with a recently published multicenter study conducted in Italy, which evaluated the impact of Nusinersen treatment on a group of adult SMA patients, including 69 individuals with type III SMA. The study reported an improvement in HRQoL over a 14-month period of Nusinersen treatment (53).

Nevertheless, it is important to note that SMA therapies are associated with high costs and uncertain outcomes, as observed in this study and others. Consequently, the Saudi Ministry of Health has engaged in outcome-based agreements with certain therapy manufacturers and implemented programs for SMA patients, which include specific eligibility criteria for accessing these therapies. However, critics of these agreements raise concerns regarding prolonged negotiation periods between manufacturers and payers, which delays access to therapies, strict eligibility criteria that may restrict access for patients who could potentially benefit, as well as potential information bias in assessing various outcomes, data documentation, and governance issues (54, 55). Additionally, the compatibility of these financial agreements with existing laws and regulations in Saudi Arabia has been questioned (56). Therefore, it is crucial to invest in health technology infrastructure, promote increased information sharing and transparency between payers and manufacturers, and reform governance and procurement practices. These steps are necessary to accommodate the rapid pace of innovation witnessed in the field of orphan drugs for rare and ultra-rare diseases (57).

Finally, the impact of premarital screening and newborn screening, along with patient support programs and caregiver training and education, should be examined to effectively address SMA in Saudi Arabia. Creating effective patient support programs that address the identified needs of the patients' and their caregivers in this study are instrumental in providing comprehensive care and enhancing patients' and their families' quality of life. Equally important is caregiver training and education, equipping caregivers with the necessary knowledge and skills to deliver optimal care and effectively navigate the challenges associated with SMA. By implementing these measures improved outcomes for individuals with SMA can be achieved in Saudi Arabia.

5 Limitations

The present study has several limitations that should be considered when interpreting the findings. First, due to its cross-sectional design, the establishment of causal relationships is not possible. Additionally, the utilization of snowball sampling introduces information bias and limits the generalizability of the findings. Therefore, future studies should implement multi-stage random probability sampling method to improve both the internal and external validity of the findings. Moreover, the reliance on proxy respondents for interviews increases the risk of information bias, as highlighted in previous studies (58). Furthermore, the potential presence of interviewer bias cannot be ruled out, which may have further contributed to information bias. It should be noted that certain questionnaire items, particularly those pertaining to the quality of sleep among caregivers, exhibited a high non-response rate. Consequently, comparisons of sleep quality across caregivers of SMA patients are constrained by the significant number of unanswered questions, which hinders comprehensive analysis. Moreover, the non-response rate across multiple questionnaire items introduces additional information bias. The presence of acquiescence bias cannot be discounted as well. Lastly, it is important to acknowledge that the assessment of quality of life was conducted using a generic scale instead of a disease-specific scale, which may limit the precision of the results (59).

6 Conclusions

Our study indicates that the characteristics of SMA and its subtypes in Saudi Arabia are comparable to those observed in other countries. SMA type I remains the most severe variant, warranting increased attention from healthcare providers and policymakers. Fortunately, the emotional burden on caregivers remains minimal, largely due to the cultural norms established in the country. On the other hand, financial expenditures, while significant, do not correlate with the severity of the disease due to the variable rates of survival. The findings of this study underscore the pressing need to improve societal awareness regarding SMA and its catastrophic consequences, particularly in light of the high rates of consanguineous marriages in Saudi Arabia. To address this issue, the implementation of public awareness campaigns, premarital screening, and newborn screening programs is strongly recommended (60–62). Additionally, publishing the treatment outcomes of SMA patients enrolled in different outcome-based payment programs (OBP) is vital for evaluating the true value of these expensive therapies. Moreover, it is imperative to design and offer various patient-support programs to address the specific needs of both patients and caregivers (63). Future studies with larger sample sizes and more robust analyses should be conducted to examine the direct medical costs and socioeconomic burden of SMA. This research will provide policymakers with valuable insights to develop preventative policies aimed at reducing the incidence of the disease, such as premarital screening.

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 humans were approved by Institutional Review Board (IRB) of King Saud University Medical City (Approval of Research Project No. E-22-6955), located in Riyadh, Saudi Arabia. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed

consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

KA: Data curation, Investigation, Project administration, Writing – original draft. MAls: Data curation, Project administration, Resources, Writing – review & editing. KA-E: Data curation, Methodology, Project administration, Writing – review & editing. AB: Methodology, Project administration, Resources, Supervision, Writing – review & editing. AM: Methodology, Resources, Writing – review & editing. AA: Resources, Supervision, Writing – review & editing. HA-H: Resources, Supervision, Writing – review & editing. SA: Resources, Supervision, Writing – review & editing. FA: Conceptualization, Methodology, Resources, Writing – review & editing. M-HT: Conceptualization, Methodology, Writing – review & editing. NA: Methodology, Project administration, Writing – review & editing. MAlw: Data curation, Methodology, Writing – review & editing. YAs: Project administration, Supervision, Writing – review & editing. YAl: Conceptualization, Formal analysis, Funding acquisition, Investigation, Project administration, Supervision, Writing – review & editing.

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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.

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Research on the risk governance of fraudulent reimbursement of patient consultation fees

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Background: The fundamental medical insurance fund, often referred to as the public's "life-saving fund," plays a crucial role in both individual well-being and the pursuit of social justice. Medicare fraudulent claims reduce "life-saving money" to "Tang's monk meat", undermining social justice and affecting social stability.

Methods: We utilized crawler technology to gather textual data from 215 cases involving fraudulent health insurance claims. Simultaneously, statistical data spanning 2018 to 2021 was collected from the official websites of the China Medical Insurance Bureau and Anhui Medical Insurance Bureau. The collected data underwent comprehensive analysis through Excel, SPSS 26.0 and R4.2.1. Differential Auto-Regressive Moving Average Model (ARIMA (p, d, q)) was used to fit the fund safety forecast model, and test the predictive validity of the forecast model on the fund security data from July 2021 to October 2023 (the fund security data of Anhui Province from September 2021 to October 2023).

Results: The outcomes revealed that fraudulent claims by health insurance stakeholders adversely impact the equity of health insurance funds. Furthermore, the risk management practices of Medicare fund administrators influence the publication of fraudulent claims cases. Notably, differences among Medicare stakeholders were observed in the prevalence of fraudulent claims. Additionally, effective governance of fraudulent claims risks was found to have a positive impact on the overall health of healthcare funds. Moreover, the predictive validity of the forecast model on the national and Anhui province's fund security data was 92.86% and 100% respectively.

Conclusion: We propose four recommendations for the governance of health insurance fraudulent claims risk behaviors. These recommendations include strategies such as "combatting health insurance fraudulent claims to preserve the fairness of health insurance funds", "introducing initiatives for fraud risk governance and strengthening awareness of the rule of law", "focusing on designated medical institutions and establishing a robust long-term regulatory system", and "adapting to contemporary needs while maintaining a focus on long-term regulation".

KEYWORDS

health insurance, fraudulent claims, behavior, risk governance, auto-regressive moving average model

Introduction

General Secretary Xi Jinping underscored in his 19th National Congress report that the increasing demand from the people for social justice and the asymmetry in supply and demand resulting from unbalanced development have emerged as the primary factors influencing current social conflicts. The conflict between the supply and demand of social health resources is particularly acute, and the impoverishment of families due to diseases has become an urgent societal issue. To a certain extent, medical insurance can prevent participants from falling into poverty or returning to destitution due to illness. However, various stakeholders in medical insurance, including participants, designated medical institutions, pharmacies, and medical insurance management, are widely involved (1), and fraudulent claims occur during the operation of medical insurance funds. As a fundamental component of social security, the management of fraudulent claims risk behavior in medical insurance, directly related to the fairness of policy implementation, is of great concern to the people and has a profound impact on social stability. Fraudulent claims transpire when the conditions for Medicare reimbursement are not met (2). In light of this, fraudulent health insurance claims can be defined as intentional and illicit actions by individuals or groups seeking compensation from health insurance funds through deception, concealment, or the provision of improper information (3, 4). These actions encompass participants falsifying medical information for health insurance reimbursement, pharmacies exploiting the sales of non-cataloged products for insurance benefits, designated medical institutions leveraging health insurance contracts for gains, and the misappropriation of funds within the management of health insurance funds. Fraud has the potential to lead to the improper disbursement of health insurance funds, the wasteful use of taxpayer dollars, and contributes to unreasonable cost-sharing within health insurance. Such fraudulent activities starkly contradict the original intent of the health insurance system and represent a pivotal element undermining its fairness in policy implementation. The risk of fraudulent claims involves the potential for engaging in deceptive practices during the claims process. Mitigating the risk of fraudulent health insurance claims is crucial for strengthening the health insurance fund system. This entails closing loopholes in fund utilization and management, assuming supervisory responsibilities, and represents the most direct approach to preventing poverty resulting from illness or a relapse into poverty. Current research on governing fraudulent claims risk primarily focuses on fraud risk analysis, identification, and anti-fraud measures. In 1963, Arrow first explored the challenges of health insurance fund regulation in the absence of doctor-patient information asymmetry and a health care cost constraint mechanism from an economic perspective (5). In the 1970s, information asymmetry theory gained traction in the insurance industry, leading to the rapid development of insurance fraud theory (1, 6, 7). In 1996, the United States, through the Health Insurance Portability and Accountability Act, officially defined health insurance fraud as knowingly implementing or attempting to implement a scheme to obtain payment from a health insurance fund through misrepresentation or deceptive promises (3). Subsequently, the most scrutinized theoretical studies (8–10) have been conducted in the interest of insurers to provide theoretical support to prevent fraudulent insurance activities. Most of the insurance fraud supervision in the UK is outside the criminal justice system, and the police play a major role in dealing with insurance fraud. Button (11) guaranteed the development of the insurance industry from the perspective of strengthening police supervision. King (12) summarized the practical experience of anti-fraud in medical insurance in the United States, and pointed out that the government should speed up the anti-fraud legislation in health insurance, strengthen the anti-fraud struggle, establish the anti-fraud data system, and improve the level of anti-fraud technology. Stiernstedt (13) proposed to establish an international private health insurance sector in preventing fraud and providing healthcare services. Thaifur (14) used game theory to analyze the conditions and processes of the game between medical institutions and basic medical insurance organizations, and reveal the causes and the necessity of the existence of moral hazard. In fraud risk identification, the emphasis is mainly on the application of data mining and neural network techniques. For example, Marisa (15) proposed a data mining approach to identify health care fund regulatory risks and used it for medical claims screening in the United States. Yang (16) constructed a fund regulatory risk identification model based on data mining techniques and conducted a practical study in Taiwan, China. Zhang (17) identified the of health insurance fraud under the payment mode of a single disease based on regional chain and deep learning methods. Ortega (18) developed a neural network anti-fraud system that identifies health insurance providers, physicians, and enrollees as subjects. Supervision and risk control of medical insurance funds mainly focus on two aspects: risk sources and risk management. Risk sources include excessive demand on the demand side (19) and induced demand on the supply side (20), reflecting moral risk. Most research on the regulation of fraudulent claims of insurance funds based on moral hazard relies on big data analysis, especially in the field of motor insurance claims, yielding positive outcomes in developing fraud detection and risk measurement methods (21) and exploring the trend of fraudulent data in claims (22). However, research on Medicare fraudulent claims risk behavior in China is relatively scarce.

We construct a conceptual model of health insurance fraudulent claims risk governance by referring to the Smith model theory (four dimensions influence public policy implementation outcomes including idealized policy, target group, implementing agency, and policy implementation environment) (23), as shown in Figure 1. The target group in the theory refers to health insurance stakeholders; the implementing agency refers to the Medicare fund manager, referred to as Offices, and the policy implementation environment refers to the

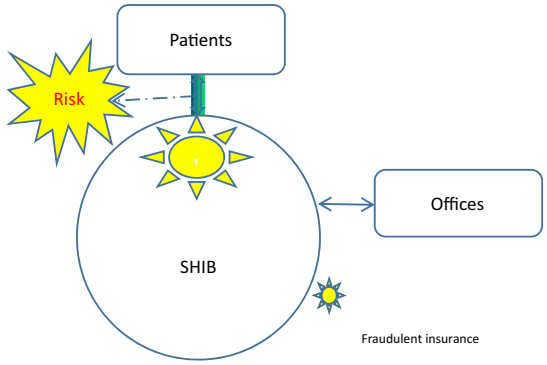

FIGURE 1
Concept model. SHIB: Stakeholders of health insurance benefits.

risk governance behavior. Based on the above-mentioned literature studies, we propose the following research hypotheses:

> *Hypothesis 1*: Fraudulent claims by Medicare stakeholders influence the equity of the Medicare fund.
>
> *Hypothesis 2*: Disparities exist among Medicare stakeholders regarding fraudulent claims in the Medicare Fund.
>
> *Hypothesis 3*: The fraudulent claims risk governance behavior of Medicare fund managers has an impact on the publication of fraudulent claims cases.
>
> *Hypothesis 4*: Positive effects are observed in the health of the Medicare fund due to fraudulent claims risk governance.

Subjects and methods

Utilizing Python for data extraction, we systematically gathered information on fraudulent insurance activities involving college students and reported cases from BaiDu from 2015 to 2021, comprehensively collected the cases reported by the National Medical Insurance Bureau and Anhui Provincial Medical Insurance Bureau from 2018 to 2021. A total of 235 cases of typical fraudulent insurance activities were initially collected, acknowledging some instances with incomplete information regarding fraudulent insurance amounts and event years. Subsequently, we carefully processed the data, resulting in the retention of 215 valid cases. The data encompassed details related to medical insurance, maternity insurance, and policy texts, sourced from the National Health Insurance Bureau and the Anhui Provincial Health Insurance Bureau.

Data extraction

Official regulatory measures for fraudulent claims encompass the establishment of a regulatory department (earning 10 points) and assigning 5 points for the involved agency. Furthermore, 1 point each is awarded for introducing a national-level document and organizing a national-level regulatory meeting. Notably, 50% of the value of the national-level document is allocated for each task involving monitoring information shared by online media. To bolster the security of the Medicare fund, a strategic allocation plan is implemented. Specifically, the Medicare security line is set at 90% of the nodal fund revenue, with the remaining 10% designated as a contingency fund. This contingency fund serves as a safety net for settling lagged costs related to out-of-province medical claims, aligning with the approach used for COVID-19 contingency risk payments. Maintaining the accuracy and reliability of data is crucial. Two researchers independently assessed the data, reaching a consensus on all aspects. Rigorous cross-verification using a double-entry method was employed to ensure error-free data entry, upholding the integrity and precision of the information.

Methods

Excel was employed to model the data distribution based on the sample data, as outlined by Sun (24). This analysis aimed to investigate the distribution of medical insurance fund stakeholders, national maternity insurance expenditure and the differences in the risk subjects of fraudulent claims of medical insurance fund. Furthermore, we analyze the income and expenditure of national medical insurance fund at different times, and Anhui Province too. SPSS26.0 was utilized to delve into the binary correlation between the regulatory behavior and fund balance of fraudulent insurance claims risk governance with the number of fraudulent insurance announcements (25–28). Based on the differential Auto-Regressive Moving Average Model (ARIMA (p, d, q)), we use the R4.2.1 software to fit the fund safety forecast model and test the predictive validity of the forecast model for fund security. Statistically significant differences were considered when $p < 0.05$.

Results

Stakeholders in the medical insurance fund

The data of medical insurance fund purchase was collected from the official website of China Medical Insurance Bureau, and the development trend of the number of people buying medical insurance is shown in Figure 2.

Figure 2 indicates that the coverage of basic medical insurance in China exceeded 1.34 billion people from 2018 to 2020. During this period, the coverage rate increased from 96.41 to 97.63% in 2018.

National maternity insurance expenditure

Considering that maternity insurance expenditure is not only related to medical insurance income, but also a part of medical insurance expenditure. Therefore, it is necessary to explore the current situation of the national maternity insurance expenditure. Figure 3 was obtained by collecting the official data.

Analysis of national maternity insurance expenditure data from January 2019 to June 2021 indicates a consistent downward trend each year. However, there is a notable exception during the period when the maternity policy is introduced.

The subject difference of fraudulent insurance pathways

We gathered 215 representative cases of online fraudulent insurance schemes from 2012 to 2021. Through case data extraction and text analysis, we obtained the corresponding percentages of subjects involved in fraudulent insurance activities (refer to Figure 4).

The study revealed that designated medical institutions accounted for 71.16% of the total cases, with a total of 153 cases. The cumulative amount involved in these cases reached 322,125,800 yuan. Although cases involving fraudulent activities by medical insurance government authorities were limited to 4, they had the highest total amount involved. Notably, the Bayannur City Social Insurance Administration irregularly used funds, leading to an amount of 10,312,000 yuan. The instances and total amounts related to fraudulent activities by designated medical institutions surpassed those of other groups.

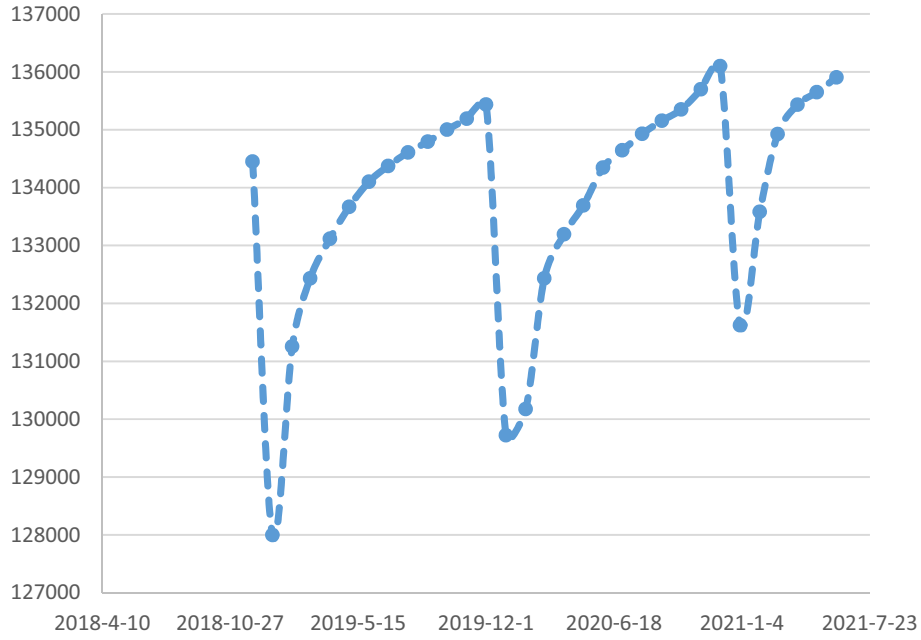

FIGURE 2
Trends in the number of people purchasing health insurance (Ten thousand).

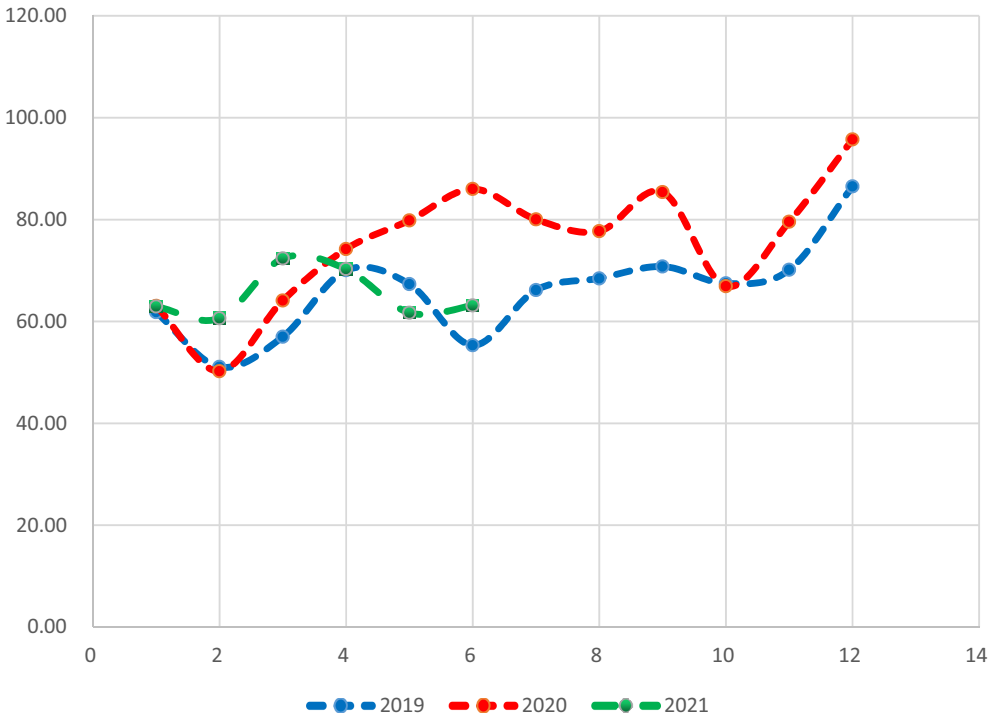

FIGURE 3
State spending on maternity insurance benefits (¥:billion).

Trends in medicare fund revenues and expenditures

Initially, we delve into the balance of national health insurance fund income and expenditure using statistical data from the National Health Insurance Administration of China. The study outcomes are depicted in Figure 5.

In the experimental group, the income of state basic health insurance (2069.718 ± 404.413) exceeds its expenditure (1733.633 ± 306.480). This difference is statistically significant (p<0.001). Additionally, the national

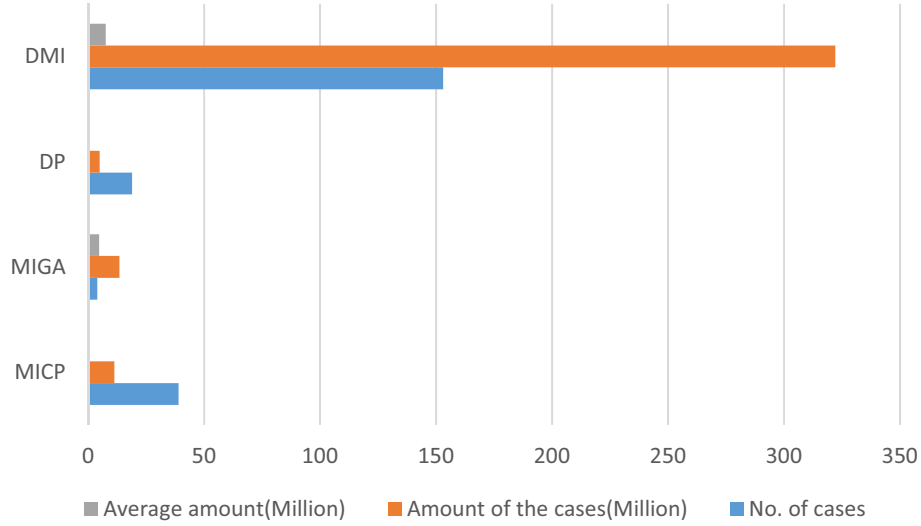

FIGURE 4
status of insurance fraud. DMI, Designated medical institutions; DP, Designated pharmacies; MIGA, Medical insurance government authorities; MICP, Medical Insurance Covered Persons.

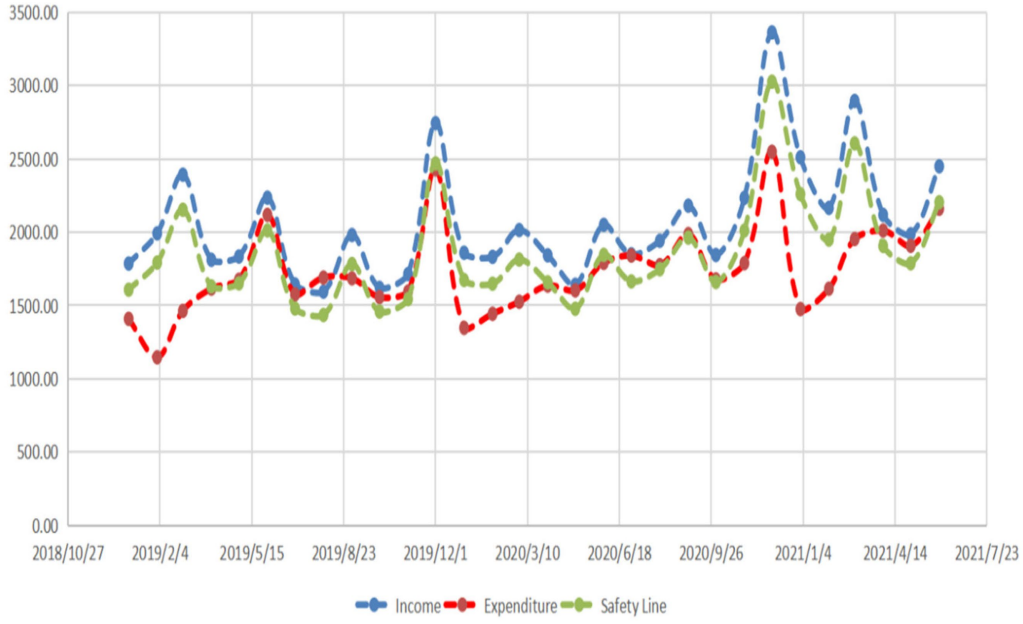

FIGURE 5
Current status of national basic medical insurance income and expenditure.

basic health insurance expenditure in the experimental group (1733.633 ± 306.480) is lower than the safety line of the national basic health insurance expenditure (1862.746 ± 363.970), with statistical significance ($p = 0.018$). We further investigate the balance of income and expenditure of the Anhui Provincial medical insurance fund, and the study results are illustrated in Figure 6.

When comparing the income (68.361 ± 38.156) and expenditure (62.585 ± 9.742) of basic medical insurance in the experimental group in Anhui, the difference was not significant ($p = 0.461$). Similarly, when comparing the safety line of basic medical insurance expenditure (62.585 ± 9.742) with the actual basic medical insurance expenditure in Anhui Province in the experimental group (61.525 ± 34.341), the difference was not statistically significant ($p = 0.881$).

From Figures 5, 6 provincial disparities in the current status of basic medical insurance income and expenditure are evident.

Binary correlation analysis

We conducted a more in-depth analysis, delving into the number of fraudulent cases, the regulatory behavior of risk management, and the annual balance amount of the Medicare fund in the context of

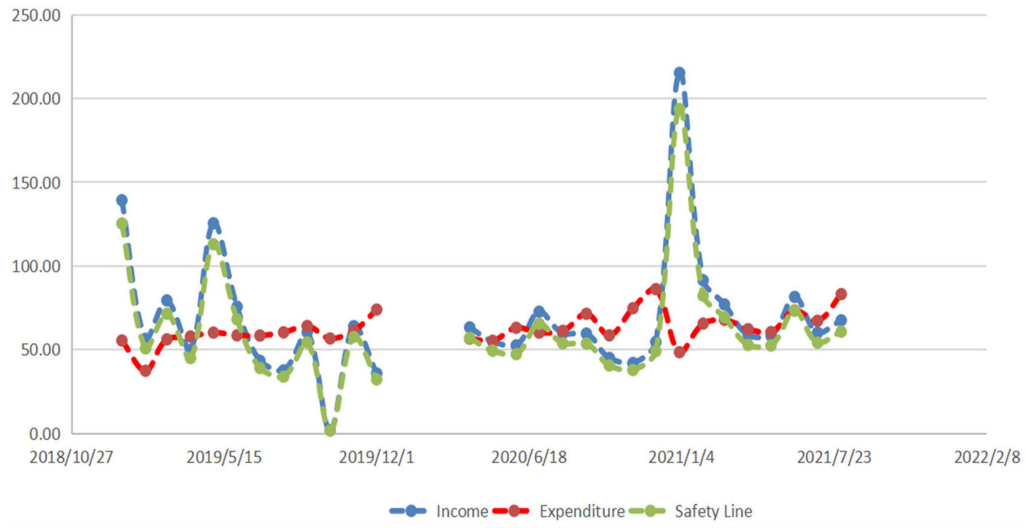

FIGURE 6
Current status of income and expenditure of basic medical insurance in Anhui Province.

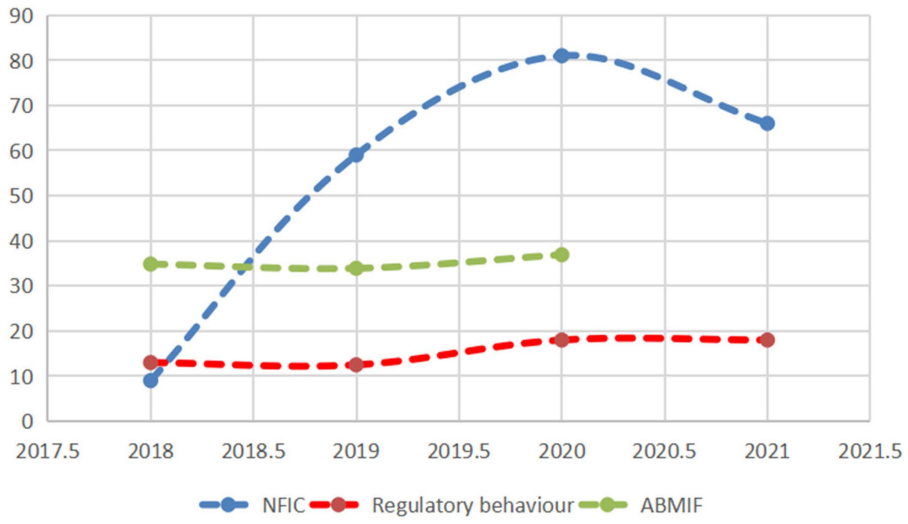

FIGURE 7
Effectiveness of official fraud and fraudulent insurance regulation. NFIC, Number of fraudulent insurance cases; ABMIF, The amount of the balance of the Medical Insurance Fund (Ten billion).

Medicare fraudulent claims risk management. The study outcomes are depicted in Figure 7.

From Figure 7, we can find that fraudulent claims risk governance positively affects the amount of annual balance of the Medicare fund. We further analyzed the correlation between the regulatory behavior and fund balance of fraudulent insurance claims risk governance with the number of fraudulent insurance announcements, and it was found that the regulatory behavior was positively correlated with the number of fraudulent insurance announcements ($r = 0.685$), the regulatory behavior was positively correlated with the fund balance ($r = 0.975$), and the fund balance was positively correlated with the number of fraudulent insurance announcements ($r = 0.495$).

Model forecast

We used the ARIMA (p, d, q), fitted the fund safety forecast model with the data of Anhui Provincial Medical Security Fund, and obtained the corresponding forecast model results of the safety line of medical insurance fund, as shown in Figures 8, 9.

In order to further verify the effectiveness of the forecast model, we used the data of the National Medical Fund from July 2021 to October 2023 (the fund security data of Anhui Province from September 2021 to October 2023) to test the predictive validity of the forecast model on the fund safety, and obtained the forecast results shown in Figures 10, 11.

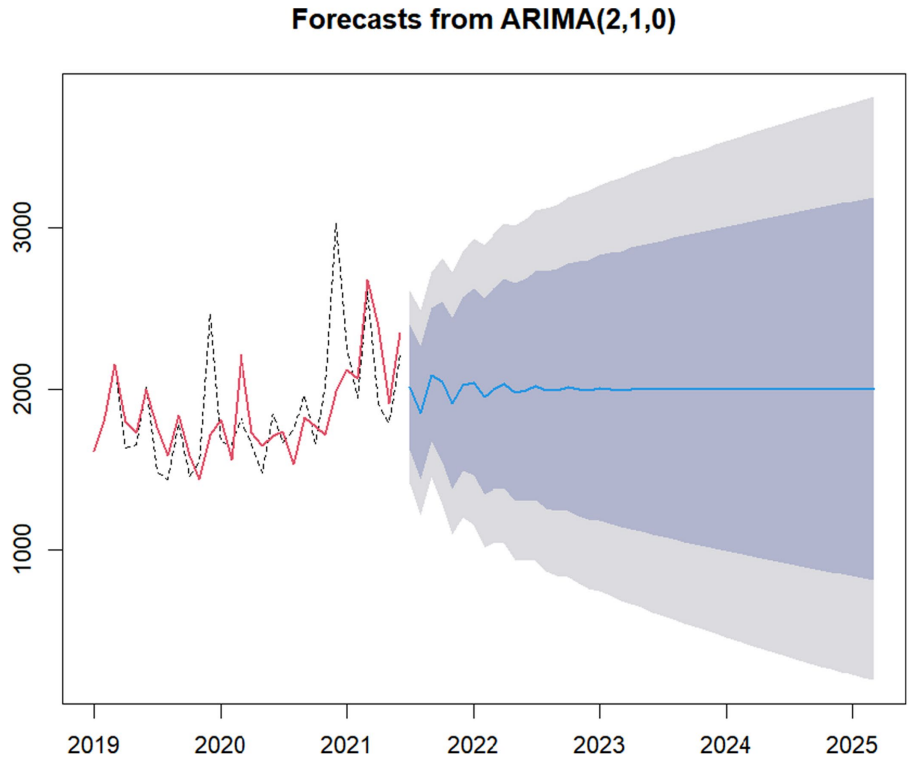

FIGURE 8
Forecasts from ARIMA (1, 1, 0) of China. The light blue area is the 95% confidence interval of the safety forecast line, the gray area is the 85% confidence interval of the safety forecast line.

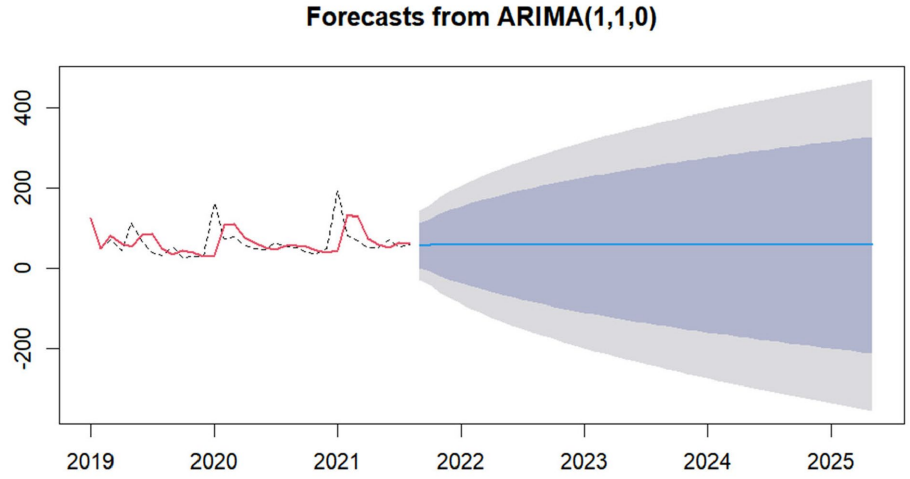

FIGURE 9
Forecasts from ARIMA (1, 1, 0) of Anhui. The light blue area is the 95% confidence interval of the safety forecast line, the gray area is the 85% confidence interval of the safety forecast line.

As easily seen in Figure 10, 28 confirmatory samples have a matching rate as high as 92.86%.

From Figure 11 easily see, with 25 confirmatory sample data, the matching rate is up to 100%.

In a word, it is not difficult to find that our fitted models ARIMA (2, 1, 0) and ARIMA (1, 1, 0) are effective.

Discussion

The findings of this study indicate that basic medical insurance in China has achieved comprehensive coverage. While a scientific and reasonable health insurance reimbursement policy can ensure equitable opportunities for health care coverage, achieving fair

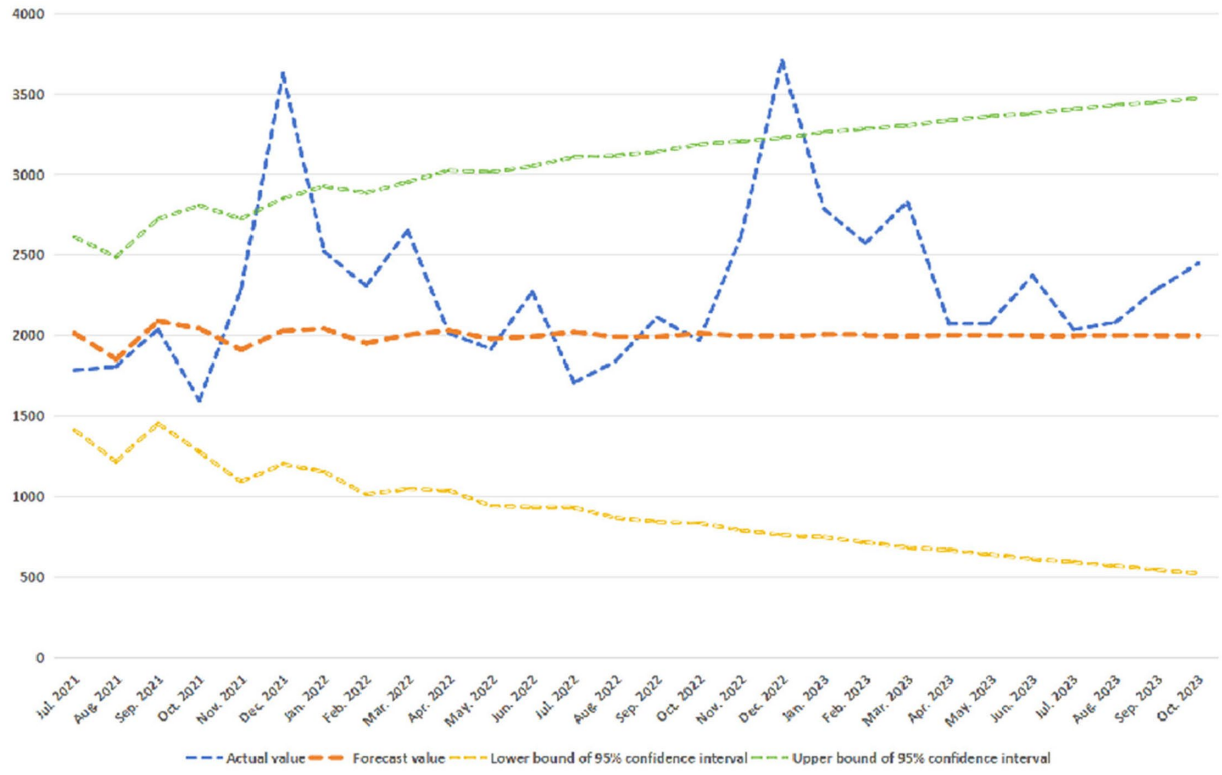

FIGURE 10
Verification of model predictive validity of China.

FIGURE 11
Verification of model predictive validity of Anhui.

outcomes depends largely on the effectiveness of the policy implementation process (20). The fixed total annual amount of the medical insurance fund for each purchaser makes it inherently unfair to ordinary consumers if fraudulent claims occur during its utilization. This, to a certain extent, underscores that fraudulent claims by medical insurance stakeholders impact the fairness of the medical insurance fund and supports Hypothesis 1. This is particularly crucial as China enters an aging era and experiences a declining fertility rate, emphasizing the direct connection between ensuring the fairness of basic medical insurance and the overall health development of China.

The study findings reveal that the highest number of insurance fraud cases and the highest total amount of cases involve medical institutions. Several possible reasons contribute to this phenomenon. Firstly, designated medical institutions, being major consumers of medical insurance funds, may lack up-to-date or clear information about the medical reimbursement system, resulting in uninformed wrongful payments. Secondly, some hospitals, under the pressure of survival, may resort to fraudulent insurance practices such as excessive medical treatment, fake hospitalization, and fictitious medical treatment items. Thirdly, the low professionalism of some physicians may lead them to prioritize financial gain over their ethical duties, prescribing "large prescriptions" and engaging in transitional examinations in collaboration with medical device companies. Fourthly, patients seeking financial gain may actively participate in hospitalization schemes offering incentives like "diet for hospitalization" and "subsidies for hospitalization," leading to the over-treatment of minor illnesses (29). Additionally, fixed-point pharmacies, restricted by the supervision of the medical security fund, engage in visual management of the network, effectively combating fraudulent practices like "food instead of drugs" and blocking pharmacies' attempts at "fraudulent insurance fraud." The fifth reason is the national health insurance fund earmarking policy, which timely cleans up the "fund misappropriation" by the main administrators of the health insurance fund, preventing irregular uses of funds such as the case of Bayannur City Social Insurance Administration, which misused funds amounting to 10,312,000 yuan. As a participant's fraudulent claim is inherently random, the main administrator of the Medicare fund significantly reduces fraudulent claims through real-name authentication and mid-way inspections. The analysis above to a certain extent confirms Hypothesis 2, indicating differences in Medicare fund fraudulent claims among health insurance stakeholder subjects.

The study results demonstrate a positive correlation between the regulatory behavior of fraudulent insurance claim risk management and the number of fraudulent insurance announcements. Several potential reasons contribute to this correlation. Firstly, since the establishment of the National Health Security Administration and the creation of the Medicare Fund Supervision Division, there has been enhanced oversight for fraud regulation within the Medicare Fund. Secondly, the aging population and low fertility rate have intensified the strain on the health insurance fund, heightening the risk to the safety of the basic health insurance fund and necessitating urgent measures to curb fraudulent practices. The third possible reason is the issuance of the "Guidance on Advancing the Reform of the Medical Security Fund Supervision System" by the General Office of the State Council on June 30, 2020, which accelerated the reform of the medical insurance fund supervision system and provided a framework for combating fraudulent insurance practices. Furthermore, the implementation of the "Regulations on Supervision and Administration of Medical Insurance Funds" on May 1, 2021, offered legal support to counter "fraudulent insurance." Specifically, according to Shi Zihai, deputy director of the National Health Insurance Bureau, inspections conducted in 2019 and 2020 revealed significant irregularities in designated pharmaceutical and medical institutions, leading to corrective actions, recoveries, fines, and a substantial recovery of the medical insurance fund. In 2021, ongoing inspections resulted in the handling of illegal institutions and the recovery of medical insurance funds and default money. Investigations into cases of "three fakes" (fake patients, fake conditions, and fake bills) also contributed to recovering misappropriated funds. This set of results underscores the effectiveness of the state's efforts in combating Medicare fund fraud and managing the risks associated with fraudulent claims, thus confirming Hypothesis 3.

The study results indicate that China's basic medical insurance fund is operating within a safe range, with a low risk of bottoming out. However, there are provincial variations in the security risk of the basic medical insurance fund. The positive impact of medical insurance fraudulent claims governance on fund security is confirmed, supporting Hypothesis 4. According to the statistical snapshot, the total revenue of the fund in 2019 was 2.4 trillion yuan, with expenditure around 2.1 trillion yuan. The current-year balance stood at 270 billion yuan, and the rollover balance from previous years exceeded 3 trillion yuan. The national basic medical insurance income was higher than the expenditure, and the expenditure was below the safety line, indicating overall smooth fund operation and abundant balance. This balance usage does not adversely affect the public's current health insurance treatment. Nevertheless, there is a less evident difference between basic medical insurance income and expenditure in Anhui Province. The basic medical insurance expenditure in Anhui is higher than the safety line, suggesting a less optimistic outlook for the health insurance fund balance. A potential explanation is the negative growth in the newborn population in Anhui from 2017 to 2020, with a significant decline in annual births. The analysis of Medicare fraud cases, risk management regulatory behaviors, and the annual balance of the Medicare Fund indicates that risk management of fraudulent claims has a positive impact on the fund's annual balance. One possible reason is that such risk management can mitigate large-scale risk losses, increasing the fund's rollover balance and safeguarding fairness in emergency responses to risk events. Additionally, risk management serves as a deterrent, minimizing fraudulent claims in the Medicare fund and enhancing fund efficiency, aligning with management philosophies found in the literature (30, 31). Other contributing factors may also play a role.

The ARIMA (1, 1, 0) and ARIMA (2, 1, 0) proposed in this study have high reliability, mainly because of the stability of our data.

Conclusion

Basic health insurance fraud poses a global governance challenge. Since the onset of COVID-19, the Chinese government's strategy of fully reimbursing health insurance for treating COVID-19 patients at public expense has led to a rapid depletion of health insurance funds, resulting in a swift decline in the "rollover" balance of the national health insurance fund. To navigate the risks associated with Medicare payments, cost-cutting measures

and expanded insurance coverage have been considered, such as increasing individual health insurance costs, delaying retirement, or extending coverage to retirees. These approaches, while addressing financial pressures, come with their own set of challenges. Examining potential cost-cutting avenues, it becomes evident that standardizing the health insurance reimbursement system (an ongoing project with continuous optimization but no ultimate goal) and recovering funds lost to fraudulent insurance activities are crucial. The emphasis should be on fund recovery and preventing financial leaks through enhanced standardized management. Building on the findings of our study, we propose four risk management pathways along with corresponding recommendations. To combat fraudulent health insurance claims and uphold fairness in health insurance funds, it is crucial to recognize that deceptive claims by medical insurance stakeholders significantly impact the equity of medical insurance funds. This underscores the pressing need to proactively manage the risks associated with fraudulent claims in health insurance. As economic growth transitions from high speed to medium speed, the aging population continues to grow, the disease spectrum evolves, and medical technology advances. Consequently, medical expenses are on a continual rise, and the situation where the growth rate of fund revenue is lower than the growth rate of expenditure is becoming the new normal. This places a certain level of pressure on the medium and long-term balance of the medical insurance fund. Effective governance of fraudulent insurance is not only a financial imperative but also crucial for maintaining social stability. Enhance fraud risk management initiatives and reinforce adherence to the rule of law. Implementing robust fraudulent claims risk management practices by administrators of the Medicare fund can lead to increased disclosure of fraudulent cases and more substantial recovery of fund losses. We recommend that national health insurance administrators actively adopt risk management initiatives, with a specific emphasis on legal frameworks. Increased investment in introducing adjudication standards is crucial to provide technical support for identifying fraudulent claims, enhancing risk control, and mitigating management risks. For instance, establishing a statutory "definition of connotation" and a clear jurisprudential basis is essential for adjudicating specific fraudulent insurance cases. Additionally, in the realm of big data screening, potential risks may arise during administrative reviews where the use of "big data screening to detect irregularities and quantify the amount of irregularities" could face challenges from local legal offices citing issues such as "lack of individual screening judgment, unclear facts, and insufficient evidence" to alter the judgment. Moreover, the construction of a credit supervision mechanism lags, with missing standards for credit supervision evaluation. The implementation lacks a strong foundation in the rule of law, such as the absence of legal basis for the hospital or health insurance fund authorities to establish a "blacklist" system for individuals with credit default. Balancing the "equilibrium point" between safeguarding individual rights to medical treatment and regulating credit default is challenging, particularly in cases where the "life first" guideline does not necessarily apply to ordinary contract disputes (economic disputes). Establishing a fund supervision system centered around fixed-point medical institutions is crucial for effectively regulating fraudulent claims within the Medicare Fund, particularly in relation to health insurance-related subjects. To enhance fraudulent claims risk management in designated medical institutions, the following recommendations are proposed: Initially, enhance agreement management by refining model agreements to align with regional specifics and resolving issues related to unclear rights and responsibilities. Actively construct credit supervision evaluation and related standards, ensuring improved efficiency and the expansion of scientific supervision channels. Additionally, promote standardization by implementing various measures such as establishing a drug standard library, defining treatment sites, and tailoring practices to match the regional context. Introduce pertinent documents and rules, propose standards for risk case identification, and develop methods for managing non-compliant data to establish a legal foundation for fund supervision. Moreover, bolster financial investment through the scientific implementation of a funding-sharing mechanism for intelligent monitoring equipment. If necessary, consider introducing social capital to facilitate the establishment of an intelligent monitoring mechanism. Implement big data screening, real-time monitoring, and timely warning systems to provide crucial technical support. The completion of these tasks will lead to standardized management, streamlined operations, and comprehensive supervision.

Staying abreast of contemporary needs and ensuring sustained regulation, the affirmative influence of governing fraudulent claims risk contributes to the well-being of health insurance funds. Tailored to the specific variations among provinces, we advocate establishing a series of systems with unique characteristics, encompassing "supervision and inspection, intelligent monitoring, credit supervision, comprehensive supervision, social supervision, and supervision and assessment." This approach seeks commonalities while respecting differences, focusing on breakthroughs, and advancing the construction of a long-term mechanism for the supervision of the entire process, cycle, and chain of medical insurance funds. This aims to safeguard the security of medical insurance funds and ensure the sustainable development of the basic medical insurance fund system. The formation of a rule of law as a means, with credit management as the foundation, and a diverse range of inspections, big data supervision as the basis for the new regulatory paradigm, will establish a comprehensive regulatory mechanism covering the entire process, cycle, and chain.

Limitations

The application of fund security forecast model to dig deeper, if further build fund income (insured quantity and mark) and quantitative relationship of fund spending, mining medical insurance fund balance internal correlation formula, can increase the dynamic balance regulation operability, this is our team need further research direction in the future. Future research could also explore perceptible changes or patterns in the medical insurance fund's operational data by using identify the organizational size, years of operation, and regional variations in operation as control variables. Alternatively, the study organizations or agencies could be stratified by these characteristics when there are detectable variations or patterns identified from the data.

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

JS: Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Validation, Writing – original draft, Writing – review & editing. YW: Formal analysis, Validation, Writing – review & editing, Writing – original draft. YZ: -. LL: Data curation, Formal analysis, Investigation, Validation, Writing – review & editing, Software. HL: Data curation, Investigation, Validation, Writing – review & editing, Formal analysis. TL: Data curation, Investigation, Validation, Writing – review & editing, Software. LZ: Formal analysis, Validation, Writing – original draft, Writing – review & editing, Data curation, Funding acquisition, Investigation, Project administration, Resources.

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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.

Publisher's note

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Application of telemedicine system for older adults postoperative patients in community: a feasibility study

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Purpose: In response to the growing challenges posed by an aging society, a telemedicine system was developed specifically for older adults postoperative patients, and its effectiveness was thoroughly investigated.

Methods: Between May 2020 and May 2022, a total of 88 older adults postoperative patients were enrolled and randomly allocated into an experimental group and a control group. The experimental group received telemedicine services after discharge, while the control group received conventional medical services following the traditional protocol. One month after discharge, various indicators were evaluated for both groups, including number of visits, medical expenditures, postoperative recovery, anxiety, depression and satisfaction.

Results: The number of visits and medical expenditures of the experimental group were less than those of the control group [1 (0, 1) vs. 1 (1, 2), $Z = -3.977$, $p < 0.001$; 25.25 (0.00, 277.40) yuan vs. 174.65 (49.63, 446.10) yuan, $Z = -2.150$, $p = 0.032$]. In both groups, there were 2 cases of incision infection, respectively. No significant difference was observed between the two groups (Fisher χ^2, $p = 0.259$). In both groups, there was no instance of incision bleeding, incision dehiscence, readmission, or reoperation. Additionally, there was no significant difference in physical status between the two groups at discharge and after discharge (66.06 ± 8.92 vs. 65.45 ± 7.39 t = 0.287, $p = 0.775$; 73.33 ± 9.97 vs. 70.91 ± 7.50, $t = 1.202$, $p = 0.235$). And there was no significant difference in the change of physical status between the two groups after discharge [10.00 (0.00, 10.00) vs. 5.00 (0.00, 10.00), $Z = -1.077$, $p = 0.281$]. There was no significant difference in body weight change between the two groups after discharge [1.05 (0.38, 1.60) Kg vs. 0.80 (0.50, 1.43) Kg, $Z = -0.265$, $p = 0.791$]. There was no significant difference in the levels of anxiety and depression between the two groups at discharge (45.64 ± 8.10 vs. 44.60 ± 8.24, $t = 0.520$, $p = 0.604$, 48.33 ± 8.46 vs. 47.50 ± 6.85, $t = 0.418$, $p = 0.677$). But the levels of anxiety and depression in the experimental group were lower than those in the control group after discharge (34.92 ± 7.38 vs. 39.03 ± 8.42, $t = -2.183$, $p = 0.032$, 37.86 ± 7.29 vs. 41.93 ± 7.13, $t = -2.281$, $p = 0.025$); The change of anxiety level and depression level of the experimental group were more than those of the control group [−10.00 (−11.25, −8.75) vs. −5.00 (−7.81, −3.75), $Z = -5.277$, $p < 0.001$; −10.00 (−12.50, −7.50) vs. −5.00 (−7.75, −3.44), $Z = -4.596$, $p < 0.001$]. The level of satisfaction regarding medical services, daily care, and psychological comfort was higher in the experimental group compared to the control group [3 (3, 3.25) vs. 2 (1, 2), $Z = -5.931$, $p < 0.001$; 3 (3, 4) vs. 3 (2, 3), $Z = -2.286$, $p = 0.022$; 2 (1, 3) vs. 1 (0.75, 2), $Z = -2.081$, $p = 0.037$].

Conclusion: In the context of an aging society, telemedicine system can offer improved healthcare to older adults postoperative patients. This includes benefits such as reducing number of visits, saving medical expenditures, enhancing psychological comfort and daily care.

KEYWORDS

aging society, postoperative, telemedicine system, telemedicine, older adults patients

1 Introduction

The population aging is accelerating at an unprecedented pace. The World Health Organization (WHO) estimates that by 2050, the proportion of the global population over 60 years will nearly double from 2015, from 12 to 22 percent (1). The process of population aging in China underwent an acceleration phase in the late 1970s and has since consistently grown at an annual rate of approximately 3.2%. Notably, this process that unfolded over more than 45 years in developed nations occurred in just about 27 years in China, and the trend of aging is expected to persist for an extended duration (2). Given the population aging, the growing demand for medical resources has created a certain contradiction between supply and demand with the existing medical resources (3).

Telemedicine, as defined by the WHO, is characterized as "healing from a distance." To be more precise, it entails the utilization of information and communication technologies to enhance patient outcomes by broadening access to medical care and information (4). A growing awareness is emerging regarding the expansive potential of remote medical care to enrich healthcare services. This approach effectively leverages limited medical resources, enhances the practices of clinical diagnosis, treatment, and disease care, and further bolsters individual health, particularly in developing nations (5). As the largest developing country, remote health services can help address China's healthcare challenges (6), in particular by alleviating the shortage of healthcare resources and the problem of centralized distribution of healthcare personnel (7). With government support and public recognition, online medical services are rapidly expanding (8).

In the present day, as society advances rapidly, adults' lives are increasingly consumed by work, especially the middle-aged face undeniable pressures. Home-based rehabilitation, caregiving, and follow-up for older adults postoperative patients are gradually witnessing a reduction in the participation of family members. Older adults individuals who have undergone surgical procedure require ongoing medical services even after being discharged home, including incision dressing change, medication management, dietary guidance, exercise recommendation, health consultation, and various other forms of postoperative care. Compounded by the fact that their children are often not medical professionals, this situation presents challenges to facilitating effective home-based rehabilitation for older adults postoperative patients. In response, our research group has developed a telemedicine system tailored for older adults postoperative patients returning home for rehabilitation. This system aims to provide enhanced convenience and high-quality medical services alongside older adults care.

2 Materials and methods

2.1 Study design

A double-blinded, randomized controlled trial was conducted from May 2020 to May 2022. Older adults patients (aged ≥65 years) who underwent surgical treatment from Wenzhou Hospital of Integrated Traditional Chinese and Western Medicine were enrolled as participants. A total of 88 participants were subsequently divided into an experimental group and a control group based on disease type, utilizing disease type as a stratification factor through a random number table in a 3:1 ratio. Due to the impact of the COVID-19 pandemic, there had been a reduction in the number of patients seeking surgical treatment, making patient recruitment challenging. Additionally, based on preliminary research indicating a favorable inclination toward remote medical care for patients, the experimental design for this study set a ratio of 3:1 between the experimental and control groups.

The implementers of the two groups were different, including a doctor and a nurse, respectively. The doctor, nurse, and patients in each group only knew the content implemented in their own group, but did not know whether they were in the experimental group or the control group, nor did they know the content implemented in the other group, nor did they know the purpose of this study.

All participants signed a written informed consent. The study protocol was approved by Ethics Committee of Wenzhou Hospital of Integrated Traditional Chinese and Western Medicine. Data and safety monitoring were handled by an independent board.

Please refer to Figure 1 for the experimental flowchart.

2.2 Inclusion and exclusion criteria

Inclusion Criteria:

1) Stable vital signs during treatment: Patients need to maintain stable vital signs during the treatment.
2) No or slight pain: Patients should not experience significant pain at discharge, or only mild pain.
3) No postoperative complications: No complication arising from surgery was found before discharge.
4) Self-feeding without intravenous nutritional support: Patients should be able to feed themselves without the need for intravenous nutritional support.
5) Normal bowel and bladder function: Postoperative patients' bowel and bladder functions should be normal.
6) Able to move: Patients should be able to get out of bed and move around.

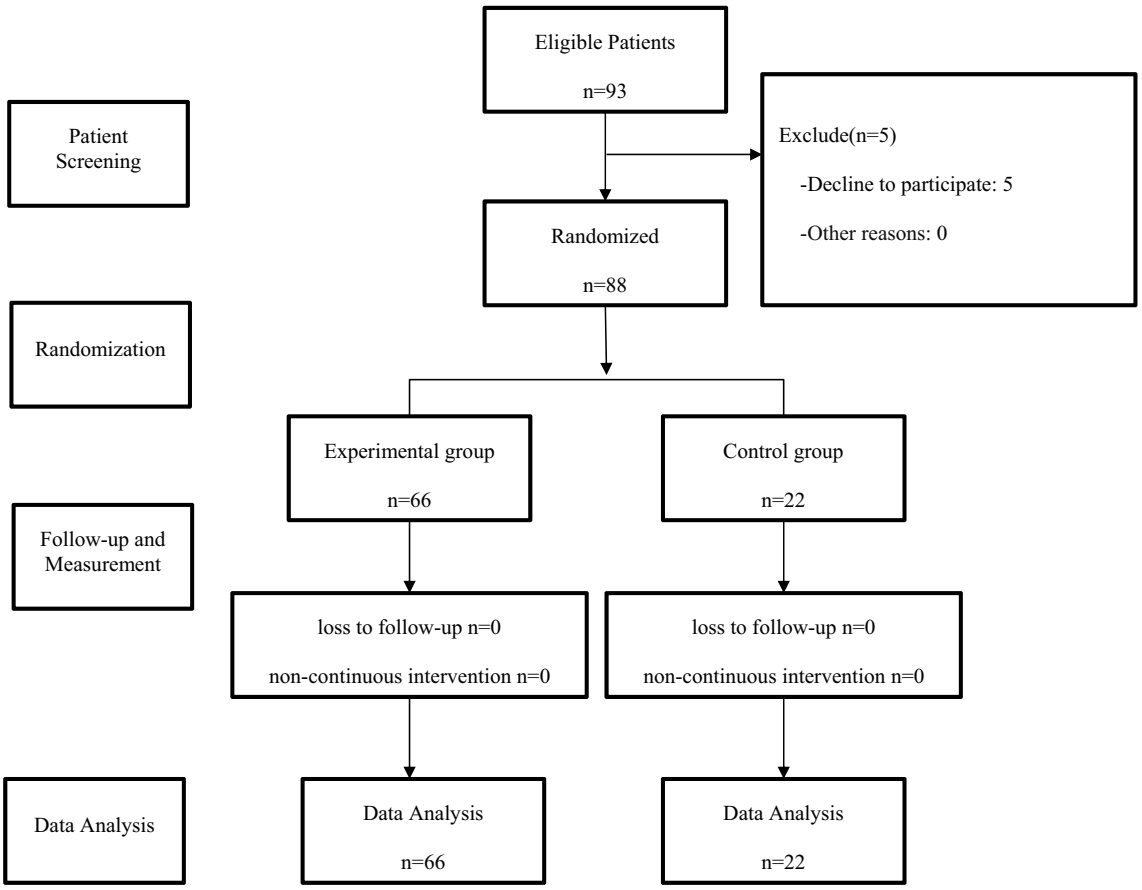

FIGURE 1
CONSORT flow diagram.

7) Well-controlled chronic diseases, stable condition: Chronic diseases of patients should have been well-controlled, and their overall health should be stable.
8) Agree to be discharged: Patients should agree to be discharged.
9) Possession of a smartphone or relevant device for video call: Patients need to have a smartphone or a relevant device capable of video call.
10) At least one accompanying family member, agreement on remote medical services by the patient and family: Patients need to have at least one accompanying family member, and both the patient and their family must agree to receive remote medical services.

Exclusion Criteria:

1) Presence of surgical contraindications: Patients with contraindications for surgery will be excluded.
2) History of mental illness for the patient or primary caregiver: Patients or their primary caregivers with a history of mental illness will be excluded from the study.

2.3 Intervention

The experimental group integrated "medical services" and "older adults care" through the utilization of remote diagnosis and treatment application and communication software such as WeChat application.

2.3.1 Pre-discharge training

Upon participants meeting the discharge criteria and expressing consent, healthcare professionals conducted basic training for their caregivers. If the primary caregiver encountered challenges in learning, another eligible family member was assigned to receive appropriate training. The training encompassed the following areas: (A) Follow-up plan. (B) Utilization of telemedicine application and communication software. (C) Guidance for changing incision dressing. (D) Medication, diet, and exercise guidance. (E) Matters needing attention. (F) Home blood pressure and/or blood sugar measurement.

2.3.2 Medical services

Healthcare professionals conducted remote consultations with patients and their families using communication software on the 3rd, 7th, and 14th day after discharge. The services encompass: (A) Collecting information about older adults patients' symptoms, as well as some basic physical signs, including mental status, consciousness, complexion, abdominal appearance, and wound healing. (B) Providing real-time guidance to caregivers during incision dressing changes. (C) Emphasizing key aspects of postoperative rehabilitation. (D) Managing chronic conditions by reviewing recent measurements such as temperature, respiration, heart rate, blood pressure, and blood sugar. Offering guidance on adjusting medication, diet, and exercise plans through remote communication software. Highlighting the significance of chronic disease management and follow-up protocol. (E) Establishing a

direct communication channel for promptly reporting of any discomfort or abnormalities in older adults postoperative patients. Simplifying hospital visits through a streamlined process, including prearranged appointments. (F) Facilitating online prescription issuance through the remote diagnosis and treatment application and communication software, followed by offline delivery of prescribed medications via Express.

2.3.3 Daily care

Personalized dietary and exercise plans were crafted to align with distinct health conditions and surgical interventions. The postoperative regimen should delineate dietary specifics, encompassing the avoidance of alcoholic beverages and spicy foods. Additionally, during the rehabilitation process, patients were reminded of relevant precautions through follow-up. Individuals who had undergone gallbladder removal were advised to moderate their intake of fatty foods to mitigate the risk of diarrhea. For patients recuperating from appendectomy or gallbladder surgery, appropriate activity was recommended to expedite the recovery of gastrointestinal functionality. Similarly, individuals undergoing surgery for great saphenous vein varicose and inguinal hernia should refrain a prolonged walk, aside from their routine activities, to avert leg swelling and hernia patch displacement. The relevant content was also presented and explained to patients and their families through text, pictures, or videos. This approach was geared toward enhancing comprehension, reinforcing information retention, and fostering adherence.

2.3.4 Psychological comfort

Throughout the follow-up process, the older adults postoperative patients received continuous encouragement and reassurance to bolster their confidence in postoperative recovery. Timely psychological counseling was provided for those displaying noticeable anxiety or depression, with psychological assistance readily available when required. Additionally, family members were encouraged to engage actively, provide attentive care, and offer patient guidance.

The control group underwent "medical services" and "older adults care" interventions according to the traditional protocol. In terms of medical services, healthcare professionals conducted telephone follow-ups with the older adults postoperative patients on the 3rd, 7th, and 14th day after discharge. The primary focus was to assess postoperative recovery and management of chronic conditions among the older adults postoperative patients. Whenever necessary, guidance was provided, and recommendations were made for hospital or local medical care. Incision dressing changes could be performed at our hospital or a local healthcare facility. Regarding "older adults care" aspects, healthcare professionals only provided some advices.

2.4 Study outcomes

Both groups underwent evaluation on the following indices. The experimental group was assessed using remote communication software, while the control group was assessed using telephone. To ensure impartiality, two nurses who were unaware of the study's methods and objectives were assigned by the research group to conduct the evaluation.

2.4.1 Number of medical visits and medical expenditures

One month after discharge, the number of medical visits and associated medical expenditures were analyzed. The count encompassed the number of visits each older adults person made to medical facilities, along with the computation of the medical expenditures incurred during these visits.

2.4.2 Postoperative recovery

One month after discharge, the occurrence of incision infection, incision bleeding, incision dehiscence, readmission, and reoperation was quantified in both groups. Additionally, physical status was assessed using the Karnofsky Performance Scale (KPS) (9) at discharge and one month after discharge, and body weight change was calculated by subtracting the weight at discharge from the weight one month after discharge.

2.4.3 Anxiety and depression

At discharge and one month after discharge, the Zung Self-Rating Anxiety Scale (SAS) (10) and Self-Rating Depression Scale (SDS) (11) were employed to assess the level of anxiety and depression of each older adults postoperative patient.

2.4.4 Satisfaction

One month after discharge, the satisfaction of the older adults postoperative patients was evaluated. The evaluation encompassed three facets: medical services, daily care, and psychological comfort. Each facet was evaluated using the Likert 5-point grading method (12, 13), encompassing responses from "very dissatisfied," "dissatisfied," "neutral," "satisfied," to "very satisfied," each assigned a score ranging from 0 to 4 points.

2.5 Statistical analysis

All study data were analyzed using SPSS 18.0 software. The *t*-test was employed to compare means in normally distributed data between the two groups. The Pearson Chi-square test, Fisher Chi-square test, or Fisher–Freeman–Halton Chi-square test was used to assess differences in count data between the two groups. For rank data and non-normally distributed data, the Mann–Whitney U rank-sum test was utilized for comparison. The two-sided tests were used in this study and a significance level of $p < 0.05$ was considered statistically significant.

3 Results

3.1 Characteristics of the participants

The experimental group comprised 66 participants, of which 36 (54.5%) were males and 30 (45.5%) were females, with an average age of 71.29 ± 5.20 years. In terms of educational background, there were 10 (15.2%) participants with high school education, 21 (31.8%) with middle school education, and 35 (53.0%) with primary school education or lower. Among these participants, 27 (40.9%) underwent inguinal hernia surgery, 15 (22.7%) had varicose great saphenous vein

TABLE 1 Basic information of the two groups.

	Experimental group (n = 66)	Control group (n = 22)	$\chi^2/t/Z$	p-value	95% CI lower limit/upper limit
Gender[#]					
Male	36 (54.5%)	13 (59.1%)	0.138	0.710	
Female	30 (45.5%)	9 (40.9%)			
Age (years)*	71.29 ± 5.20	72.09 ± 5.03	−0.632	0.529	−3.328/1.722
Education level[†]					
Primary school	35 (53.0%)	13 (59.1%)	−0.456	0.648	
Junior high school	21 (31.8%)	6 (27.3%)			
High school and above	10 (15.2%)	3 (13.6%)			
Type of operation[¤]					
Inguinal hernia	27 (40.9%)	9 (40.9%)	0.347	1.000	
Great saphenous vein	15 (22.7%)	5 (22.7%)			
Appendix	15 (22.7%)	5 (22.7%)			
Gall bladder	9 (13.6%)	3 (13.6%)			
Chronic disease[¤]					
High blood pressure	15 (22.7%)	7 (31.8%)	1.067	0.801	
Diabetes	10 (15.2%)	3 (13.6%)			
Else	8 (12.1%)	3 (13.6%)			
None	33 (50.0%)	9 (40.9%)			

Data are presented as numbers with percentages (%) or mean ± standard deviation.
[#]Categorical variables were compared using Person chi-square test.
[¤]Categorical variables were compared using Fisher–Freeman–Halton chi-square test.
*Normally distributed data were compared using t-test.
[†]Rank data were compared using the Mann–Whitney U rank sum test.

TABLE 2 Number of visits and medical expenditures of the two groups.

	Experimental group (n = 66)	Control group (n = 22)	$/Z$	p-value
Number of visits	1 (0, 1)	1 (1, 2)	−3.977	<0.001
Medical expenditures (yuan)	25.25 (0.00, 277.40)	174.65 (49.63, 446.10)	−2.150	0.032

Data are presented as median and quartile (P25, P75).
Non-normally distributed data were compared using the Mann–Whitney U rank sum test.

surgery, 15 (22.7%) had appendix surgery, and 9 (13.6%) underwent gallbladder surgery. In terms of past history, it included 15 (22.7%) cases of hypertension, 10 (15.2%) cases of diabetes mellitus and 8 (12.1%) cases of other chronic diseases. In the control group, there were 22 participants, including 13 (59.1%) males and 9 (40.9%) females, with an average age of 72.09 ± 5.03 years. In terms of educational background, there were 3 (13.6%) participants with high school education, 6 (27.3%) with middle school education, and 13 (59.1%) with primary school education and lower. Among them, 9 (40.9%) underwent inguinal hernia surgery, 5 (22.7%) had varicose great saphenous vein surgery, 5 (22.7%) had appendix surgery, and 3 (13.6%) underwent gallbladder surgery. Moreover, the control group consisted of 7 (31.8%) patients with hypertension, 3 (13.6%) with diabetes, and 3 (13.6%) with other chronic illnesses. Notably, there was no significant difference in basic demographic characteristics between the two groups ($p > 0.05$). For detailed information, please refer to Table 1.

3.2 Medical visits and medical expenditures

The number of visits of the experimental group was 1 (0, 1), whereas that of the control group was 1 (1, 2). The difference between the two groups was statistically significant ($Z = -3.977$, $p < 0.001$).

The medical expenditures of the experimental group amounted to 25.25 (0.00, 277.40) yuan, whereas that of the control group amounted to 174.65 (49.63, 446.10) yuan. The difference between the two groups was statistically significant ($Z = -2.150$, $p = 0.032$) (see Table 2 for details).

3.3 Postoperative recovery

Two cases of incision infection occurred in each of the two groups, and there was no statistically significant difference between the two groups (Fisher χ^2, $p = 0.259$). Notably, no instance of incision bleeding,

TABLE 3 Physical status and body weight change of the two groups.

	Experimental group (n = 66)	Control group (n = 22)	t/Z	p-value	95% CI lower limit/upper limit
Physical status at-discharge*	66.06 ± 8.92	65.45 ± 7.39	0.287	0.775	−3.590/4.801
Physical status post-discharge*	73.33 ± 9.97	70.91 ± 7.50	1.202	0.235	−1.630/6.479
Change of physical status#	10.00 (0.00, 10.00)	5.00 (0.00, 10.00)	−1.077	0.281	
Body weight change (Kg)#	1.05 (0.38, 1.60)	0.80 (0.50, 1.43)	−0.265	0.791	–

Data are presented as mean ± standard deviation or median and quartile (P25, P75).
The change of physical status is calculated by subtracting the physical status score at discharge from the physical status score after discharge.
*Normally distributed data were compared using t-test.
#Non-normally distributed data were compared using the Mann–Whitney U rank sum test.

TABLE 4 Anxiety and depression of the two groups.

	Experimental group (n = 66)	Control group (n = 22)	t/Z	p-value	95% CI lower limit/upper limit
Anxiety at discharge*	45.64 ± 8.10	44.60 ± 8.24	0.520	0.604	−2.941/5.024
Anxiety after discharge*	34.92 ± 7.38	39.03 ± 8.42	−2.183	0.032	−7.853/−0.367
Change of anxiety#	−10.00 (−11.25, −8.75)	−5.00 (−7.81, −3.75)	−5.277	<0.001	
Depression at discharge*	48.33 ± 8.46	47.50 ± 6.85	0.418	0.677	−3.128/4.795
Depression after discharge*	37.86 ± 7.29	41.93 ± 7.13	−2.281	0.025	−7.620/−0.524
Change of depression#	−10.00 (−12.50, −7.50)	−5.00 (−7.75, −3.44)	−4.596	<0.001	

Data are presented as mean ± standard deviation or median and quartile (P25, P75).
The Change of anxiety/depression is calculated by subtracting the anxiety/ depression score at discharge from the anxiety/depression score after discharge.
*Normally distributed data were compared using t-test.
#Non-normally distributed data were compared using the Mann–Whitney U rank sum test.

incision dehiscence, readmission, or reoperation was reported in either of the two groups. There was no significant difference in physical status between the two groups at discharge and after discharge (66.06 ± 8.92 vs. 65.45 ± 7.39, t = 0.287, p = 0.775; 73.33 ± 9.97 vs. 70.91 ± 7.50, t = 1.202, p = 0.235). And there was no significant difference in the change of physical status between the two groups after discharge [10.00 (0.00, 10.00) vs. 5.00 (0.00, 10.00), Z = −1.077, p = 0.281]. Additionally, there was no significant difference in body weight change between the two groups after discharge [1.05 (0.38, 1.60) Kg vs. 0.80 (0.50, 1.43) Kg, Z = −0.265, p = 0.791] (see Table 3 for details).

3.4 Anxiety and depression

There was no significant difference in the levels of anxiety and depression between the two groups at discharge (45.64 ± 8.10 vs. 44.60 ± 8.24, t = 0.520, p = 0.604; 48.33 ± 8.46 vs. 47.50 ± 6.85, t = 0.418, p = 0.677). But the levels of anxiety and depression in the experimental group were lower than those in the control group after discharge (34.92 ± 7.38 vs. 39.03 ± 8.42, t = −2.183, p = 0.032; 37.86 ± 7.29 vs. 41.93 ± 7.13, t = −2.281, p = 0.025); The change of Anxiety level and Depression level of the experimental group were more than those of the control group [−10.00 (−11.25, −8.75) vs. −5.00(−7.81, −3.75), Z = −5.277, p < 0.001; −10.00 (−12.50, −7.50)vs. −5.00(−7.75, −3.44), Z = −4.596, p < 0.001] (see Table 4 for details).

3.5 Patient satisfaction

The level of satisfaction regarding medical services, daily care, and psychological comfort was higher in the experimental group compared to the control group [3 (3, 3.25) vs. 2 (1,2), Z = −5.931, p < 0.001; 3 (3, 4) vs. 3 (2, 3), Z = −2.286, p = 0.022; 2 (1, 3) vs. 1 (0.75, 2), Z = −2.081, p = 0.037] (see Table 5 for details).

4 Discussion

4.1 The telemedicine system can better meet the medical requirements of older adults postoperative patients

The study revealed that older adults postoperative patients in the experimental group made fewer hospital visits compared to those in the control group, resulting in lower medical costs. This indicates that the remote medical care model is more effective in addressing the medical requirements of older adults postoperative patients.

TABLE 5 Satisfaction level of the two groups.

	Experimental group (n = 66)	Control group (n = 22)	Z	p-value
Medical services	3 (3, 3.25)	2 (1, 2)	−5.931	<0.001
Daily care	3 (3, 4)	3 (2, 3)	−2.286	0.022
Psychological comfort	2 (1, 3)	1 (0.75, 2)	−2.081	0.037

Data are presented as median and quartile (P25, P75).
Non-normally distributed data were compared using the Mann–Whitney U rank sum test.

Based on a systematic review, an individual's social, cultural, and digital technological competence is a crucial factor in determining the benefits derived from telemedical care (14). Consequently, this study diligently endeavored to impart suitable training to patients and their families enrolled in the telemedicine system. Caregivers were ultimately empowered and assist older adults in changing incision dressings independently or under the guidance of a telemedicine specialist.

When an older adults individual was feeling unwell, a medical professional can conduct a remote assessment. Certain symptoms could be addressed through temporary observation, modifications to diet/exercise, or suitable medication, effectively eliminating the necessity for a hospital visit. This approach served to alleviate patient anxiety and also helps in minimizing unnecessary visits. As for the no difference in recovery between the two groups, this might be related to the insufficient sample size.

About 65% of the older adults suffered from various chronic diseases (15), such as hypertension (16) and diabetes (17, 18). Consequently, while the telemedicine system devised in this study primarily targeted the enhancement of rehabilitation among the older adults postoperative patients, it also encompassed an indispensable facet of chronic disease management. The study revealed instances where blood pressure and blood sugar levels of certain chronic patients in the control group were inadequately managed after discharge. In contrast, the experimental group benefited from the guidance provided by medical professionals, resulting in more favorable outcomes. A study demonstrated that regular Blood Pressure Tracking (BPT), compared to conventional treatment, could significantly lower blood pressure, particularly for high-risk hypertensive patients (19, 20). Coincidentally, remote medical care was also advantageous for controlling blood sugar levels in diabetic patients (21). In the telemedicine system established in this study, well-trained caregivers played an important role in routinely monitoring the blood pressure and blood sugar of the older adults.

Additionally, this service model provided convenience for medical staff to adjust treatment and care plans in a timely manner based on changes in patients' conditions, such as providing remote guidance or supervision in areas such as diet, exercise, and medication treatment. The study by Barbosa et al. (22) also indicated that satisfactory treatment outcomes could be achieved through network-enabled remote management of chronic diseases. Generally, by effectively utilizing smart devices and the WeChat platform, it was possible to further enhance the accessibility of medical services and the quality of healthcare for surgical patients and their families. This aligned with the findings of previous researches (23–25).

4.2 The telemedicine system can better meet the psychological requirements of older adults postoperative patients

The occurrence of COVID-19 pandemic could lead to anxiety, depression, and other adverse emotions among the older adults. Particularly, older adults who had undergone surgical treatment were more susceptible to experiencing anxiety and depression (26). This psychological vulnerability arised from concerns not only about postoperative rehabilitation but also the management of chronic diseases. In instances where a patient experienced excessive worry about their condition, medical personnel could address the concerns of the older adults through an objective assessment of the situation. In our actual research, we discovered that the older adults postoperative patients, to varying degrees, tended to experience concerns about the successfulness of their recovery following discharge. These concerns often revolved around issues such as medication adjustment plan, time for follow-up appointment, dietary restriction, optimal level of physical activity, and permissibility of bathing. Even after receiving answers to these recurring queries, they might still raise the same question again. Such a scenario was likely indicative of anxiety. An experiment involving remote medical care for older adults patients indicated that among the participants receiving remote medical care, 36% of patients required essential technical assistance, while 58% of patients needed additional psychological support due to a lack of confidence (27).

Within this system, caregivers of the older adults underwent fundamental training. This equipped them with an understanding of the older adult's condition and post-discharge care requirements, enabling them to offer explanations to the older adults, replacing the need for repeated involvement of medical staff. Furthermore, video calls between healthcare professionals and the older adults within this system might be more effective in enhancing patients' psychological well-being than telephone calls in the control group. For instance, medical staff could distinctly perceive the expressions of the older adults and foster a stronger sense of trust between them, consequently facilitating medical staff to provide more effective psychological counseling for the older adults. Similarly, Lim et al. (28) also discovered in their research on remote chronic disease management that this approach could effectively alleviate anxiety and depression among older adults patients. This finding corresponded with the conclusions drawn from our study. This indicated that the widespread adoption of mobile technology could alleviate patients' psychological health needs (29).

4.3 The telemedicine system can improve the satisfaction of older adults postoperative patients

The study revealed that the older adults postoperative patients in the experimental group exhibited higher levels of satisfaction with medical services, daily care, and psychological comfort compared to those in the control group. This observation suggested that the telemedicine system had a more favorable impact in these particular areas.

4.3.1 Medical services

Collaborative efforts between doctors and nurses within the telemedical system resulted in improved the effectiveness of postoperative rehabilitation guidance and enhanced chronic disease management for the older adults. This, in turn, leaded to heightened satisfaction with medical services among the older adults. A retrospective survey indicated that telemedicine was found satisfactory on various outcome and the most common advantages were time saved (30).

4.3.2 Daily care

Within the context of the telemedicine system, family members engaged in remote follow-up, enabling them to gain a deeper understanding of the older adults postoperative patients' physical and mental well-being, and subsequently provided meticulous care. Additionally, the remote follow-up by medical staff also served as a catalyst for motivating family members, thereby potentially leading to more standardized and refined daily care.

4.3.3 Psychological comfort

The telemedicine system harmonized the involvement of medical staff and family members in the postoperative management of the older adults. This collaborative approach helped to gain better insight into the psychological states of the older adults. When necessary, prompt psychological counseling could be offered, resulting in increased psychological comfort for the older adults. Furthermore, the combined effect of enhanced daily care and improved medical services can contribute to alleviating anxiety and depression among older adults individuals. Existing research indicated that mobile technology-based applications not only facilitated familial connectivity but also established a connection between the older adults and healthcare resources, fostering enhancements in both physical and mental well-being (21).

In this study, as all patients were older adults, the scoring sheet was designed to request simple ratings directly from patients only in the aspects of medical services, daily care, and psychological comfort. Because each aspect was evaluated using a single item, we did not measure the reliability and validity of the questionnaire. To ensure the stability of the measurement, the measurements of both groups were carried out by the same personnel, and the same understandable text expression and language explanation of item was used during the measurement. In future research, adding rating items may enhance scientific validity of data.

But this study only discusses patients who can participate in telemedicine system, and for some older adults individuals, the digital divide may pose a limitation. In cases where patients are unable to access the telemedicine system due to various reasons, it is crucial to establish support measures for telemedicine system management. Firstly, design a user-friendly interface that meets the unique needs and preferences of older adults users. Additionally, provide clear and concise instructions on how to access telemedicine platforms and resources. Implementing user training courses or tutorials may further enhance digital literacy among the older adults population. Moreover, establish a dedicated support system, such as a hotline or online assistance, to address any technological challenges or concerns. By taking these measures, healthcare providers can optimize the use of telemedicine tools, improving the overall experience and outcomes for older adults postoperative patients. Proficient individuals, utilizing smart devices, can access fundamental daily care and rehabilitation procedures through the hospital's social platforms. This not only alleviates the workload of doctors but also enhances the quality of medical services (31).

5 Conclusion

In summary, the integrated telemedicine model developed in this study holds in line with the contemporary landscape of increasing aging population. This model amalgamates medical services and older adults care, facilitating older adults postoperative patients to curtail needless medical visits while experiencing top-notch healthcare provisions within community settings. This approach fosters the physical and psychological wellness of older adults postoperative patients. Consequently, advocating for its broader dissemination and adoption is justified. To foster the advancement of telemedicine, governmental agencies must enhance regulation and integration, while the healthcare system should offer support in personnel and software systems. Importantly, it is necessary to amplify public awareness and acceptance of remote medical care. The comprehensive development of telemedicine demands collaborative efforts from diverse sectors.

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 humans were approved by Wenzhou Hospital of Integrated Traditional Chinese and Western Medicine Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

Q-PW: Conceptualization, Data curation, Investigation, Writing – original draft. W-YC: Conceptualization, Data curation, Investigation, Writing – original draft. M-MH: Data curation, Formal analysis, Writing – original draft. Y-XH: Data curation, Formal analysis, Writing – original draft. S-SL: Formal analysis, Writing – review & editing. Y-CG: Formal analysis, Writing – review & editing.

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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.

Publisher's note

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Appraisal of universal health insurance and maternal health services utilization: pre- and post-context of the Jaminan Kesehatan Nasional implementation in Indonesia

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Introduction: The Indonesian government introduced universal health insurance through the National Social Security System (JKN) in 2014 to enhance overall healthcare. This study compares maternal health care (MHC) service utilization before and after JKN implementation in Indonesia.

Method: Using 2012 and 2017 data from Indonesia Demographic and Health Surveys (DHS), we conducted a two-period cross-sectional design study following the Anderson model. We assessed how the JKN policy and population characteristics influenced healthcare utilization for women aged 15–49 who had given birth in the last 5 years. Multivariable logistic regression models were used to assess the impact of the JKN policy and related factors.

Result: In two waves of Indonesia DHS with 14,782 and 15,021 subjects, this study observed a significant increase in maternal healthcare service utilization post-JKN implementation. Women were more likely to have at least four antenatal care visits (adjusted odds ratio, AOR = 1.17), receive skilled antenatal care (AOR = 1.49), obtain skilled birth assistance (AOR = 1.96), and access facility-based delivery (AOR = 2.45) compared with pre-JKN implementation.

Conclusion: This study revealed a significant positive impact of JKN on enhancing MHS utilization. The introduction of universal health insurance coverage likely reduced financial barriers for specific demographics, resulting in increased service utilization. Our study may offer valuable insights for Asian countries with similar demographics and health insurance implementations.

KEYWORDS

National Social Security System, Jaminan Kesehatan Nasional, universal health coverage, maternal care, Demographic and Health Surveys (DHS)

Introduction

More than three-quarters of the women in WHO member states receive maternal services yet the extent of intraregional inequality remains overwhelming (1). In 2020, the daily count of women succumbing to pregnancy and childbirth complications exceeded 800, with 95% of these fatalities occurring in low and lower-middle-income countries (2). Indonesia, among Southeast Asia nations, exhibits a significantly higher maternal mortality ratio, with 189 maternal deaths per 100,000 live births (3). Despite the recommendations for prenatal care, a study discovered that 91.2% of women received inadequate antenatal care, resulting in a 1.3 fold increase in the likelihood of labor complications (4). The rate of institutional childbirth rose from 22% in 1986 to 73% in 2012 (5). Nevertheless, the absence of a consistent reduction in maternal deaths stems from substandard service quality in both within primary care setting and hospitals (6, 7).

Persistent challenges in low and lower middle-income countries are linked to Sustainable Development Goals (SDGs) 3, which sets a target to decrease the global maternal mortality ratio to less than 70 per 100,000 live births by year 2030. Analogously, SDGs 5, which focuses on gender equality and women's rights, shares a comparable focus (8). In addition, the World Health Organization is committed to ensuring comprehensive maternal health services through the Ending Preventable Maternal Mortality Strategy (8). This program considers various factors, including health systems, universal health coverage (UHC), and socioeconomic determinants. It also emphasizes policy measures to support family planning, healthy pregnancies, and safe childbirth (9).

Numerous initiatives were introduced to bolster maternal health services in Indonesia, including the global Safe Motherhood Initiative was formulated during a conference in Nairobi in 1987 (10), with Indonesia holding its first national seminar on safe motherhood in 1988 (11). Subsequently, the village midwife (*bidan di desa*) program was established. Providing trained midwives in village birth facilities (*polindes*). The Action to Cherish Mother (*Gerakan Sayang Ibu*) was launched in 1996 as a government-led effort to reduce maternal mortality. Despite this intervention, traditional birth attendance (*dukun*) remains prevalent at the community level. Traditional birth attendants (dukun) typically lack formal training in modern medicine, which may result in lower service quality (12).

In 2014, the Indonesian government implemented a UHC initiative known as National Social Security System (JKN), with the core objective of offering comprehensive coverage for all citizens, including access to maternity care (13). Indonesia achieved universal maternal health coverage through a range of channels, including Askes (public sector social insurance coverage), Jamsostek (private sector social insurance coverage), and Jampersal (childbirth assurance coverage). These programs were eventually consolidated into a single-payer system under JKN, providing a comprehensive maternal health benefit package (14). Furthermore, the government introduced Jamkesmas (non-contributory social insurance coverage for the impoverished) and Jamkesda (local government-funded social insurance), specifically targeting individuals who were not covered by existing insurance schemes (12). It aims to ensure that all participants are rounded up by fair and equitable services. Every citizen is required to pay a premium of 5% of monthly earnings for both public and private sector workers. However, economically disadvantaged individuals, underprivilege, or have a disability receive ongoing healthcare premium subsidies from either central or local governments, as stipulated in Legislation Number 101 of 2013. In the early semester of 2014, JKN participants reached a number of 124.55 million people and increased to 241.79 million in 2022 (15).

The earlier examination indicated a substantial enhancement in maternal health service utilization due to the implementation of JKN (16, 17). A recent study by Aryastami and Mubasyiroh (16) utilized data from the 2018 Basic Health Research of Riskesdas and suggested that women with health insurance may be more likely to utilize maternal health services optimally. Another paper by Anindya et al. (17) assessed the impact of Indonesia's National Health Scheme on access to maternal health services. However, most of these studies relied on single-year cross-sectional data to investigate the influence of health insurance coverage on maternal health service utilization, rather than emphasizing the assessment of pre- and post-policy effects on care. Furthermore, there was a lack of information regarding the existence of variations in the utilization and readiness of maternal health services across different regions within Indonesia (14, 18).

This study utilized two waives of data from the Indonesia Demographic Health Surveys (DHS) conducted in 2012 and 2017, covering the period before and after the implementation of the universal health insurance system through the National Social Security System in Indonesia in 2014. The objective was to assess the influence of JKN on maternal health service (MHS) utilization, specifically to compare MHS utilization between the periods prior to (Phase 1 in 2012) and following (Phase 2 in 2017) the implementation of JKN. To delve further into this investigation on engaging policymakers and relevant stakeholders who can serve as conduits between various entities, stratification analyses were conducted to precisely determine the extent of MHS utilization before and after the introduction of JKN among different Indonesian women subgroups within the reproductive age group.

Materials and methods

Conceptual framework

The Anderson behavioral model was adopted as the conceptual framework in this study for investigating factors associated with maternal health services utilizations. Many literatures have used this framework developed by Ronald M. Andersen in 1968 and advanced version for investigating individual and contextual determinants of health service use (19–22). As Figure 1 shows the conceptual framework for the association between health care system (before and after the implementation of JKN in 2014), population characteristics (predisposing factors, enabling factors, and needs) and utilization of maternal health services.

Study setting, design and data source

Indonesia is an archipelagic nation situated in Southeast Asia, known as the world's largest island nation, comprising over 17,000 islands. The five primary islands are Java, Sumatra, Borneo (Kalimantan), Sulawesi, and Papua, in addition to numerous smaller islands. Among women aged 15–49 in Indonesia, traditional patriarchal culture in Indonesia still exerts its influence on the choices

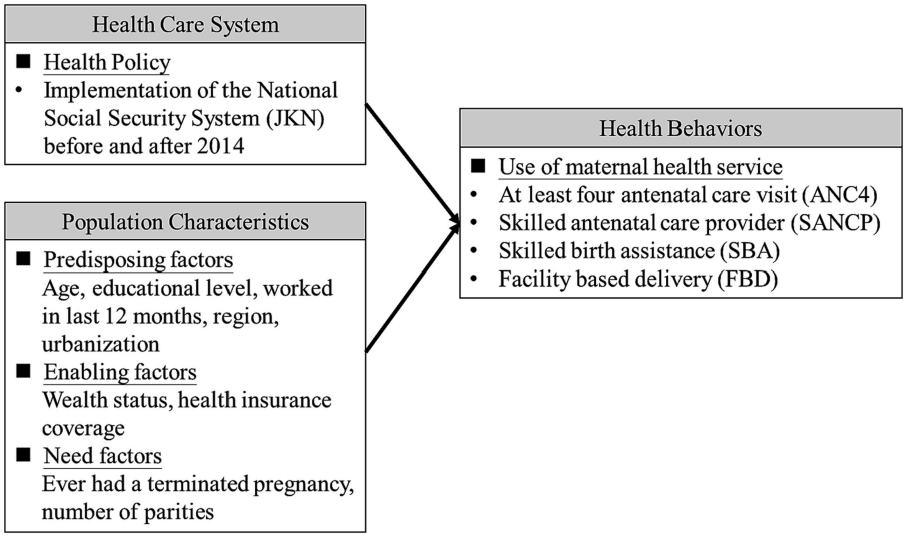

FIGURE 1
Conceptual framework of Anderson behavioral model in this study.

and behaviors of a considerable number of Indonesian women in particular respects of their lives (23). Furthermore, there exists a notable variation in the educational attainment of this demographic (24). Women of maternal age are at a higher risk of experiencing unmet healthcare needs and may be less likely to receive adequate health services in Indonesia.

This study employed a two-period cross-sectional study design, utilizing two waves of data from the Indonesia Demographic Health Surveys (DHS) conducted in 2012 and 2017. These surveys spanned both pre- and post-implementation periods, allowing for an assessment of the JKN implementation. DHS surveys are conducted in collaboration between national government agencies and international organizations, providing comprehensive data on various demographic and health indicators in developing countries, making it a valuable resource for research and evaluate healthcare demand and provision for the improvement of women and children's health. The survey datasets, accessible through an online application request for research and study purposes, have been approved by the International Coaching Federation (ICF) Institutional Review Board. Ethical clearance for the 2012 IDHS-VI and 2017 IDHS-VII was obtained from the National Institute of Health Research and Development, Indonesia Ministry of Health. Each request for data access is considered for a specific research project (25).[1]

Study population and sampling technique

The two waves of Indonesia DHS included a representative population of 43,852 and 47,963 households nationwide, including all administrative regions of the country, and both urban and rural areas, respectively in all 33 provinces in 2012 and 34 provinces in 2017. A two-stage stratification cluster design prevailed to determine a study participant, the first stage was by selecting enumeration areas (EA) from census files, and the second stage was by selecting households' samples in each EA selected to be interviewed.

This study only included women aged 15–49 who had given birth within the 5 years preceding two waves of the women's questionnaire in the Indonesia DHS survey. To streamline the analysis, a dataset was generated by consolidating all pertinent information regarding their most recent childbirth. The final number of eligible subjects for analysis was 14,782 women in 2012 (phase 1) and 15,021 in 2017 (phase 2).

Variable measurements

The outcomes of interest were MHS utilizations women aged 15 to 49 who had given birth within the 5 years in two waves of Indonesia DHS survey between the periods before (Phase 1 in 2012) and after (Phase 2 in 2017) JKN implementation. Specifically, the study included four binary variables for measuring MHS utilizations, including whether a woman received at least Four Antenatal Care (ANC4, yes/no), visited a Skilled Antenatal Care Provider (SANCP, yes/no), gave birth with Skilled Birth Assistance (SBA, yes/no), and utilized a Facility-Based Delivery (FBD, yes/no).

The primary explanatory variable of this study was the implementation of the JKN policy. The JKN implementation is categorized into pre-implementation (Phase1 in year 2012) and post-implementation (Phase 2 in year 2017). Furthermore, following the Anderson behavioral model, this study included predisposing indicators (age [<25/25–29/30–34/35–39/≥ 40], educational level [no education and primary education/ secondary education/ higher education], work status [yes/no], and regions [Java/Sumatra/ Bali and Nusa Tenggara/ Kalimantan/ Sulawesi/ Maluku and Papua], and urbanization [urban/rural]), enabling indicators (household wealth status [poor/ middle/ rich], and health insurance coverage [no covered/ covered]), and need indicators (ever had complications during pregnancy [yes/no], and number of parity [<2/>=2]).

1 http://www.measuredhs.com

Statistical analysis

This study employed weighted statistical calculations to account for population size and ensure data representativeness. Given the study's focus on female respondents, we exclusively applied the weights designated for women. In the initial phase, a new variable was created by dividing a specific women-related variable (V005) from the DHS by 1,000,000, and this variable was subsequently used in tabulations. Percentage and frequency data were used to describe population characteristics and maternal health service (MHS) utilization, comparing the periods before (Phase 1 in 2012) and after (Phase 2 in 2017) JKN implementation. We used the Chi-square test to compare differences in characteristics and outcomes attributed to the JKN policy's effects. To examine the association between the JKN policy effect and four MHS outcomes, while controlling for the population characteristics outlined in Table 1, we conducted multivariate logistic regressions. Adjusted odds ratios (AORs) and 95% confidence intervals (CIs) were reported. Furthermore, this study conducted stratified analyses to assess the JKN policy effect among various population subgroups, summarizing the results using forest plots. The study was conducted from September 2022 to July 2023. Data analysis for this research was carried out using IBM SPSS Statistics, Version 20.0, and statistical significance was considered for p-values <0.05.

Results

Table 1 shows general characteristics of women who gave births in the last 5 years. A total of 14,782 women participated in phase 1, while 15,021 women were involved in phase 2. In phase 1, women aged 25–29 years old were the most prominent ($n = 4,063$, 27.49%), whereas in phase 2, those aged 30–34 years old were the largest group ($n = 3,833$, 25.52%). The majority of women in both phases had completed secondary education ($n = 7,987$, 54.03% in phase 1; $n = 8,754$, 58.28% in phase 2), were employed in the last 12 months ($n = 7,926$, 53.62% in phase 1; $n = 7,760$, 51.66% in phase 2), resided in rural areas ($n = 7,432$, 50.28% in phase 1; $n = 7,737$, 51.51% in phase 2). Furthermore, there was an increase in health insurance coverage from phase 1 ($n = 5,364$, 36.29%) to phase 2 to ($n = 8,808$, 58.64%).

Table 2 presents the distribution of women's utilization of MHS before and after JKN implementation. There was an increase in the number of women receiving antenatal care ≥4 visits from 12,974 women (87.79%) in phase 1 with 110 missing values to 13,603 women (90.59%) in phase 2 with 65 missing values. Similarly, the proportion of respondents visiting skilled antenatal care providers increased from 14,147 women (95.70%) in phase 1 with 65 missing values to 14,647 women (97.50%) in phase 2 with 41 missing values. Additionally, the number of women giving birth with skilled birth assistance rose from 12,466 women (84.30%) in phase 1 with 60 missing values to 13,787 women (91.80%) in phase 2 with 36 missing values. Furthermore, the utilization of facility-based delivery increased from 9,541 women (64.79%) in phase 1 with 54 missing values to 12,076 women (80.59%) in phase 2 with 37 missing values. The study findings demonstrated a significant association between all maternal health services utilization and the two phases of cohorts ($p < 0.001$). Due to the small number of missing values, subjects with missing value were not considered and excluded for further analysis.

TABLE 1 Demographic characteristics of respondents in Phase 1 (2012) and Phase 2 (2017).

Parameter	Phase 1 (2012)	Phase 2 (2017)	p-value[a]
N	14,782	15,021	
Predisposing [*n* (%)]			
Age (years)			
< 25	3,355 (22.70%)	2,856 (19.01%)	<0.001
25–29	4,063 (27.49%)	3,791 (25.24%)	
30–34	3,562 (24.10%)	3,833 (25.52%)	
35–39	2,495 (16.88%)	3,010 (20.04%)	
≥ 40	1,307 (8.84%)	1,531 (10.19%)	
Educational level			
No education and primary education	5,031 (34.03%)	4,064 (27.05%)	<0.001
Secondary education	7,987 (54.03%)	8,754 (58.28%)	
Higher education	1,764 (11.94%)	2,203 (14.67%)	
Worked in last 12 months			
No	6,856 (46.38%)	7,261 (48.34%)	0.001
Yes	7,926 (53.62%)	7,760 (51.66%)	
Region			
Java	8,145 (55.10%)	8,257 (54.97%)	0.652
Sumatera	3,317 (22.44%)	3,374 (22.46%)	
Bali and Nusa Tenggara	896 (6.06%)	946 (6.30%)	
Kalimantan	925 (6.26%)	952 (6.34%)	
Sulawesi	1,078 (7.29%)	1,036 (6.90%)	
Maluku and Papua	421 (2.85%)	456 (3.04%)	
Urbanization			
Urban	7,350 (49.72%)	7,284 (48.49%)	0.034
Rural	7,432 (50.28%)	7,737 (51.51%)	
Enabling [n (%)]			
Wealth status			
Poor	5,916 (40.02%)	6,008 (39.99%)	0.220
Middle	2,939 (19.88%)	3,099 (20.63%)	
Rich	5,927 (40.10%)	5,915 (39.38%)	
Health insurance coverage			
Noncovered	9,418 (63.71%)	6,213 (41.36%)	<0.001
Covered	5,364 (36.29%)	8,808 (58.64%)	
Need [n (%)]			
Ever had a terminated pregnancy			
No	12,779 (86.45%)	12,868 (85.67%)	0.051
Yes	2,003 (13.55%)	2,153 (14.33%)	
Number of parities			
> 2	5,226 (35.35%)	5,472 (36.43%)	0.053
≤ 2	9,556 (64.65%)	9,549 (63.57%)	

[a]Chi-square test to compare differences in characteristics and outcomes attributed to the JKN policy's effects and p-value was presented.

TABLE 2 Distribution of maternal health care services utilization.

Parameter	Phase 1 (2012)	Phase 2 (2017)	*p*-value[a]
Antenatal care			
<4 visits	1,698 (11.49%)	1,353 (9.01%)	<0.001
≥4 visits	12,974 (87.79%)	13,603 (90.59%)	
Missing	110 (0.72%)	65 (0.40%)	
Skilled antenatal care provider			
No	570 (3.86%)	333 (2.22%)	<0.001
Yes	14,147 (95.70%)	14,647 (97.50%)	
Missing	65 (0.44%)	41 (0.28%)	
Skilled birth assistance			
No	2,256 (15.30%)	1,198 (8.00%)	<0.001
Yes	12,466 (84.30%)	13,787 (91.80%)	
Missing	60 (0.40%)	36 (0.20%)	
Facility based delivery			
Home	5,186 (35.21%)	2,908 (19.41%)	<0.001
Health facilities	9,541 (64.79%)	12,076 (80.59%)	
Missing	54 (0.37%)	37 (0.25%)	

[a]Chi-square test to compare differences in characteristics and outcomes attributed to the JKN policy's effects and *p*-value was presented.

Table 3 shows the multivariable logistic regression results for examining policy factors, predisposing, needs, and enabling factors associated with MHS utilization. With respect to the policy effect of the implementation of the JKN implementation (compared with Phase 1), Indonesian women were found to be more likely to have at least four antenatal care visits (aOR = 1.17, 95%CI = 1.07 ~ 1.27), received care from skilled antenatal care provider (aOR = 1.49, 95%CI = 1.29 ~ 1.73), receive skilled birth assistance (aOR = 1.96, 95%CI = 1.81 ~ 2.13), and accessed facility based delivery (aOR = 2.45, 95%CI = 2.30 ~ 2.60).

In relation to predisposing factors, as Table 3 shows, compared with reference groups, women aged 25–29, and 30–34 had a higher likelihood of utilizing ANC4, while those aged 35–39 were more likely to utilize SBA and FBD. Additionally, women with higher education levels were more inclined to utilize all four MHS services in both phases. Living in regions other than Java was associated with a reduced likelihood of utilizing all four MHS services. Moreover, residing in a rural area was linked to a reduced likelihood of accessing all four MHS services. Regarding enabling indicators, women with middle and rich wealth status and with health insurance coverage were more likely to use all four MHS services. As for need indicators, women with a history of terminated pregnancies were more likely to utilize SBA.

Figure 2 further provides a summary of the stratification results for the overall and policy factor effects on the four types of maternal health services examined in this study. These results are based on subgroups related to predisposing, enabling, and need factors. The findings indicate a significant overall increase in the likelihood of Indonesian women accessing all four MHS services after the implementation of the JKN. However, this effect may not be significant for women with higher education levels, those residing in the Sumatera region, and those living in urban areas. Additionally, the effect may not be significant for ANC4 among women with rich and middle wealth status, as well as for SANCP among women with rich wealth status.

Discussion

This study compared MHS utilization before (Phase 1 in 2012) and after (Phase 2 in 2017) the implementation of JKN. The findings revealed a significant increase in Indonesian women's likelihood to access all four MHS components: having at least four antenatal care visits, receiving care from skilled antenatal care providers, obtaining skilled birth assistance, and accessing facility-based delivery. These findings align with another research conducted in Indonesia, which also highlighted improvements in MHS utilization (26). Following the implementation of the JKN program, our results are also consistent with a study conducted across low and low middle income countries, which suggested that universal health insurance programs can have beneficial effects on MHS utilization (25, 27). The prenatal care contributes significantly to reducing maternal and neonatal morbidity and mortality, making it an essential component of comprehensive MHS. Furthermore, an increase in the number of antenatal care components received by women corresponds to a higher likelihood of them delivering at a health facility and attending postnatal care services (28).

Our study used the Anderson behavioral model as conceptual framework to access MHS utilization revealed noteworthy findings. The analysis of predisposing, enabling, and need indicators in subgroups revealed a consistent positive correlation across all indicators with a higher likelihood of utilizing MHS. In terms of predisposing indicators, specific age groups were associated with increased utilization of prenatal care. For instance, women aged 35–39 years had the highest likelihood of receiving antenatal care visits, care from skilled antenatal care providers, while women aged over 40 years had the highest likelihood of receiving skilled birth assistance and accessing facility-based delivery. This finding is consistent with earlier studies conducted in the USA and Mexico (29, 30).

The involvement of well-educated women significantly influences the choice to utilize MHS, but less impact from the implementation of the JKN program. Consistent with Laksono et al. (31) study, our results indicated the JKN program may have a prominent decrease in magnitude of socioeconomic inequality with respect to education factors. These women tend to engage in higher-paying jobs, affording them the ability to manage medical costs. Furthermore, their education empowers them with an improved understanding of their fundamental human rights and boosts their health literacy (32). As a result, it becomes crucial to improve the education system and community engagement in order to elevate the utilization of MHS (33). This study found that lower education levels had a negative impact on increasing the likelihood of utilizing multiple MHS components through the JKN program. Lower educational attainment is often associated with lower socioeconomic status, which can lead to financial constraints. With the government's initiatives to enhance maternal and child health and make healthcare more affordable, women with lower education levels may be more inclined to engage in prenatal care. This, in turn, results in a greater reliance on public health services and an increased dependence on healthcare advice from professionals (34).

TABLE 3 Results from multivariable logistic regressions to examine policy Factors, predisposing, needs, and enabling Factors associated with four types of MHS utilization.

Variables	At least four antenatal care visits		Skilled antenatal care provider		Skilled birth assistance		Facility based delivery	
	AOR(95%CI)	*p*-value	AOR (95%CI)	*p*-value	AOR(95%CI)	*p*-value	AOR (95%CI)	*p*-value
Policy factor								
Pre-JKN Implementation	1.00		1.00		1.00		1.00	
Post-JKN Implementation	1.17 (1.07,1.27)	<0.001	1.49 (1.29,1.73)	<0.001	1.96 (1.81,2.13)	<0.001	2.45 (2.30,2.60)	<0.001
Predisposing								
Age (years)								
< 25	1.00		1.00		1.00		1.00	
25–29	1.18 (1.05,1.32)	0.005	1.15 (0.93,1.41)	0.188	1.11 (0.99,1.24)	0.068	1.00 (0.92,1.09)	0.957
30–34	1.22 (1.08,1.38)	0.001	1.07 (0.87,1.33)	0.527	1.15 (1.03,1.30)	0.015	1.04 (0.95,1.14)	0.351
35–39	1.01 (0.90,1.15)	0.817	0.85 (0.69,1.06)	0.157	1.39 (1.22,1.58)	<0.001	1.21 (1.10,1.33)	<0.001
≥ 40	0.82 (0.71,0.95)	0.008	0.70 (0.55,0.90)	0.005	1.18 (1.02,1.37)	0.025	1.05 (0.93,1.18)	0.431
Educational level								
No education and primary education			1.00		1.00		1.00	
Secondary education	2.10 (1.92,2.29)	<0.001	2.97 (2.53,3.48)	<0.001	3.13 (2.88,3.41)	<0.001	2.35 (2.20,2.51)	<0.001
Higher education	3.03 (2.51,3.67)	<0.001	5.41 (3.36,8.70)	<0.001	7.52 (5.83,9.70)	<0.001	3.78 (3.33,4.29)	<0.001
Worked in last 12 months								
No	1.00		1.00		1.00		1.00	
Yes	0.93 (0.85,1.00)	0.061	0.92 (0.80,1.06)	0.249	0.82 (0.75,0.88)	<0.001	0.88 (0.83,0.93)	<0.001
Region								
Java	1.00		1.00		1.00		1.00	
Sumatera	0.42 (0.38,0.46)	<0.001	0.44 (0.36,0.53)	<0.001	1.38 (1.23,1.53)	<0.001	0.45 (0.42,0.49)	<0.001
Bali and Nusa Tenggara	1.13 (0.93,1.36)	0.222	0.82 (0.60,1.13)	0.232	0.89 (0.77,1.04)	0.132	1.21 (1.06,1.37)	0.005
Kalimantan	0.61 (0.52,0.71)	<0.001	0.39 (0.30,0.50)	<0.001	0.93 (0.80,1.09)	0.376	0.31 (0.27,0.34)	<0.001
Sulawesi	0.40 (0.35,0.45)	<0.001	0.51 (0.39,0.66)	<0.001	0.62 (0.55,0.71)	<0.001	0.34 (0.31,0.38)	<0.001
Maluku and Papua	0.17 (0.14,0.20)	<0.001	0.10 (0.08,0.12)	<0.001	0.26 (0.22,0.31)	<0.001	0.16 (0.14,0.19)	<0.001
Urbanization								
Urban	1.00		1.00		1.00		1.00	
Rural	0.91 (0.83,1.00)	0.055	0.72 (0.60,0.86)	<0.001	0.58 (0.53,0.64)	<0.001	0.42 (0.39,0.45)	<0.001
Enabling								
Wealth status								
Poor	1.00		1.00		1.00		1.00	
Middle	1.90 (1.70,2.13)	<0.001	2.50 (1.98,3.15)	<0.001	2.51 (2.24,2.81)	<0.001	1.61 (1.49,1.74)	<0.001
Rich	2.90 (2.56,3.27)	<0.001	4.60 (3.45,6.13)	<0.001	4.23 (3.71,4.83)	<0.001	2.53 (2.33,2.74)	<0.001
Health insurance coverage								
Noncovered	1.00		1.00		1.00		1.00	
Covered	1.41 (1.29,1.53)	<0.001	1.73 (1.48,2.02)	<0.001	1.27 (1.17,1.38)	<0.001	1.28 (1.20,1.36)	<0.001
Need								
Ever had a terminated pregnancy								
No	1.00		1.00		1.00		1.00	
Yes	1.13 (1.01,1.28)	0.034	1.16 (0.94,1.42)	0.168	0.96 (0.85,1.07)	0.439	1.08 (0.99,1.18)	0.087
Number of parities								
> 2	1.00		1.00		1.00		1.00	
≤ 2	0.96 (0.89,1.05)	0.393	1.11 (0.96,1.28)	0.164	1.01 (0.93,1.09)	0.824	1.03 (0.97,1.10)	0.283

AOR, adjusted odds ratio; CI, confidence interval; JKN, Jaminan Kesehatan Nasional.

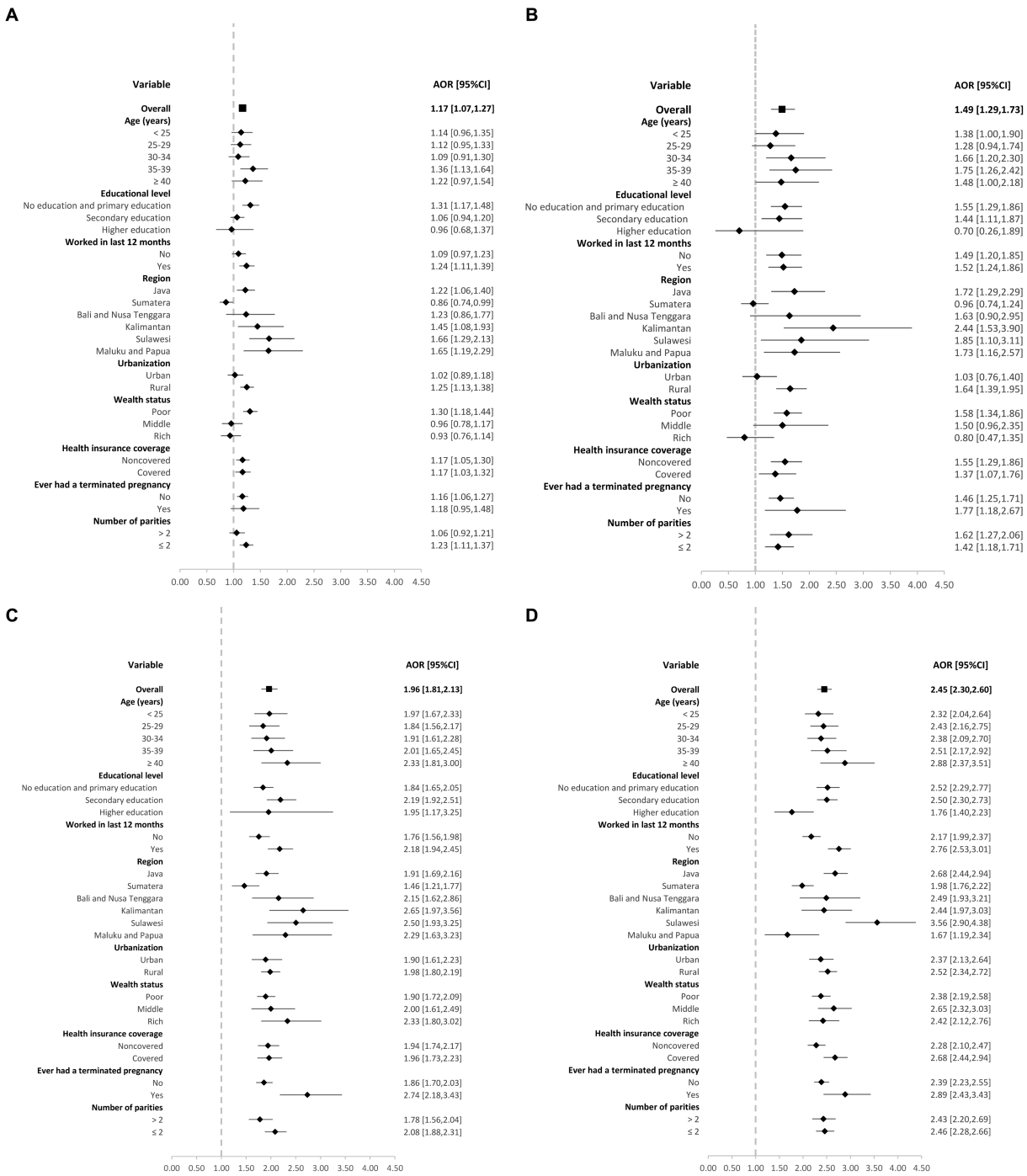

FIGURE 2
Forest plot of subgroup stratification analysis of the effect of the JKN program implementation on four maternal health services utilizationby each predisposing, enabling and need variables: **(A)** at least four antenatal care visits **(B)** skilled antenatal care provider **(C)** skilled birth assistance **(D)** facility-based delivery.

Regarding to the region factors, the government's implementation of the village midwife program, combined with the inclusion of communities already covered by regional health insurance (Jamkesda), led to a significant increase in their utilization of healthcare services (35). Despite the overall improvement in MHS utilization across different regions of Indonesia due to the implementation of JKN, the national health insurance scheme has not reached its full potential compared with Java region (36). Notably, our stratification analysis findings indicated that women residing in Sumatera experienced a lower impact on their likelihood of utilizing MHS as the results of the implementation of JKN program. A study conducted in the Sumatera region identified predisposing factors contributing to challenges in MHS utilization. These factors included the respondent's knowledge and attitude toward MHS, the influence of family members in

healthcare decisions, and community beliefs related to MHS that did not align with health-related values (37).

In terms of enabling indicators, household wealth status, and health insurance coverage status demonstrated a tendency toward a higher likelihood of utilizing MHS. The core objective of UHC predominantly centers around addressing inequalities in healthcare among the population, particularly focusing on reaching the most vulnerable group. The current study consistent with findings from Bangladesh, which indicated that women from wealthier households exhibited a propensity to utilize antenatal care (38). Poor wealth status might be linked with a greater need for healthcare services due to a higher prevalence of health risks and conditions associated with poverty. Nevertheless, our study findings affirm that women with lower household wealth index exhibited an increase in the utilization of all four MHS services after the implementation of the JKN. Furthermore, the current study found an increasing number of any types of health insurance coverage from 36.29% in 2012 to 58.64% in 2017 after the implementation of the JKN among study subjects. It may increase accessibility to financial incentives through JKN programs and might contribute to an elevated likelihood of utilizing antenatal care (31).

As to the need indicators, women who had never experienced pregnancy termination and had a parity status of less than 2 times displayed a significantly higher likelihood of utilizing MHS. Women who have never experienced a pregnancy termination may be more likely to utilize MHS due to a higher level of reproductive awareness. They may exhibit greater apprehension regarding pregnancy complications and, as a result, seek the assistance of trained healthcare professionals more diligently (39). Additionally, women with lower parity may have more time and resources at their disposal to prioritize their own health and the health of their child. In line with this, as the fourth most populous country globally, Indonesia initiated a family planning program focused on fertility reduction through a two-child policy in 1957. This program was institutionalized in 1970 with the establishment of the National for Coordination of Family Planning (*Badan Koordinasi Kelarga Berencana Nasional, BKKBN*) (40). In some countries, women encountered a financial strain when accessing MHS and are additionally required to cover certain unofficial charges, even in the presence of formal exemptions (41). This issue becomes particularly significant if they have a larger number of children, as it contributes to an escalation in the overall incurred costs.

The primary strength of this study is its extensive national dataset, which enhances its potential for generalizability and informs policymaking at the country level. However, there are limitations that should be acknowledged. First, despite the focus on the most recent births, there is a potential for recall bias, as women were required to recall events within the preceding 5 years before the survey. Second, the associations observed might be underestimated, as only surviving mothers were available for interviews, potentially excluding uninsured women who lacked antenatal and delivery care and experienced fatal childbirth complications. Third, reliance on self-reported data introduces the possibility of under- or over-reporting certain issues. Fourth, as this analysis is based on secondary data, some well-recognized predictors of service utilization, such as cultural beliefs and family factors, are absent from our evaluations. Fifth, the cross-sectional nature of the study design used makes it impossible to establish causality. Finally, the study did not collect data on healthcare usage and out-of-pocket payments, limiting the investigation of the effects of health insurance ownership.

Conclusion

This study compared maternal health service (MHS) utilization before (Phase 1 in 2012) and after (Phase 2 in 2017) the implementation of JKN, revealing a significant positive impact of JKN on enhancing MHS utilization. The introduction of universal health insurance coverage likely reduced financial barriers for specific demographics, resulting in increased service utilization. The study's stratification analysis, which includes various characteristic subgroups, provided a deeper understanding of maternal health services utilization. Our study may offer valuable insights for Asian countries with similar demographics and health insurance implementations.

Data availability statement

Publicly available datasets were analyzed in this study. This data can be found here: the DHS survey datasets, accessible through an online application request for research and study purposes, have been approved by the ICF International Institutional Review Board. Each request for data access is considered for a specific research project (http://www.measuredhs.com).

Author contributions

TR: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Software, Visualization, Writing – original draft, Writing – review & editing. H-MH: Conceptualization, Investigation, Methodology, Project administration, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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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.

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Identifying key mental health and improvement factors in hospital administrators working from home using a DEMATEL-based network analysis model

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Purpose: To identify the key mental health and improvement factors in hospital administrators working from home during COVID-19 normalization prevention and control.

Methods: The survey was conducted from May to June 2023, and the practical experiences of 33 hospital administrators were collected using purposive sampling. The study examined a set of mental health factor systems. The relationship structure between the factors was constructed using the Decision-making Trial and Evaluation Laboratory (DEMATEL) method. Finally, the structure was transformed using the influence weight of each factor via the DEMATEL-based Analytic Network Process.

Results: Regarding influence weight, the key mental health factors of hospital administrators are mainly "lack of coordination," "time management issues," and "work-life imbalances." The influential network relation map shows that improvements can be made by addressing "improper guidelines," "laziness due to being at home," and "job insecurity" because they are the main sources of influence. The reliability level of the results for the network structure and weight was 98.79% (i.e., the gap was 1.12% < 5%).

Conclusion: The network analysis model based on DEMATEL proposed in this study can evaluate the mental health factors of hospital administrators during the pandemic period from a multidimensional and multidirectional perspective and may help improve mental health problems and provide suggestions for hospital administrators.

KEYWORDS

mental health, work from home, Decision-making Trial and Evaluation Laboratory-based Analytic Network Process (DEMATEL-based ANP or DANP), multi-criteria decision-making (MCDM), COVID-19 normalization prevention and control

Introduction

The COVID-19 outbreak is a public health emergency of international concern that spread rapidly worldwide and gradually evolved into a pandemic with disastrous consequences (1, 2). COVID-19 seriously threatens people's health and global security, and has caused incalculable losses to the global economy, education, and medical care (3, 4). Doctors and nurses are at the frontline of prevention and control of the COVID-19 epidemic and play a key role in preventing infection and treating patients (5). However, during the outbreak, doctors and nurses were exhausted and understaffed, posing certain risks to public health (6). The high risk of COVID-19 infection can seriously affect the mental health of doctors and nurses, and they may be anxious about infecting other personnel (7, 8). COVID-19 is one of the main representatives of sudden major infectious diseases. Hospitals are the main institutions that fight against major infectious diseases. Therefore, the related topics concerning hospitals require investigation, especially concerning major epidemics. This includes the mental health problems of anti-epidemic roles such as doctors, nurses, and administrators need special attention.

During the fight against COVID-19, some studies focused on the mental health of doctors and nurses because they were frontline workers in the fight against the epidemic. For example, a survey in a Spanish general hospital found that more than 36% of the staff were infected with COVID-19, of whom 32% were asymptomatic (9). One study conducted a psychological survey of 9,138 medical staff and found that 45.7% of them had mental disorders, of which 14.5% were even more serious (10). In addition, one study found that 80% of confirmed patients still suffered from fatigue, cognitive impairment, dyspnea, and other sequelae after recovery (11). Doctors and nurses are important actors in the fight against COVID-19. Doctors and nurses who are infected and isolated leave the clinical front line, which causes a shortage of pandemic prevention personnel and increases the workload of other colleagues (12). Simultaneously, they worry they will infect their families, relatives, and neighbors (13). The above indications show that a shortage of personnel, self-isolation, illness, and death of confirmed patients all cause an emotional burden on doctors and nurses (14). With the epidemic changing from confrontation transformation to normal prevention and control, office and study environments have shifted online. Therefore, people who work or study at home also merit attention.

During the epidemic, to reduce cross-infection in hospitals and reduce the ability to prevent and control the epidemic, hospitals advocate for non-major medical or nursing posts to work at home, among which administrative staff are the main group working at home (15). According to one survey, most administrators who work from home, such as medical staff, also experience mental health problems (16). However, hospital administrators, as the main employees working at home, engage in many complicated and tedious administrative tasks. Their mental health has also been seriously affected; however, little attention has been paid to this issue. Therefore, it is necessary to study mental health problems faced by hospital administrators working at home (17). To address this research gap, it is necessary to analyze the key factors of hospital administrators' mental health to serve as a reference for the mental health management of hospital administrators during the potential major infectious epidemic in the future.

Mental health is usually evaluated from multiple factors/dimensions, which are suitable for multi-criteria decision-making (MCDM) as an analytical method. Moreover, the Decision-making Trial and Evaluation Laboratory-based Analytic Network Process (DEMATEL-based DANP or DANP) method can establish an influence relationship structure diagram and assess influence weights. The influence relationship structure diagram can help decision-makers understand the interaction between all factors (18). The influence weights can help decision-makers identify key factors in the system (19).

Materials and methods

Research design and analysis process

To understand the impact of homework on the mental health of hospital administrators during a major epidemic, this study quantitatively transforms the practical experience of hospital administrators into numerical values. It visualizes the relationship structure and corresponding influence weights of their psychological factors. Hospital policymakers can distinguish the degree of interaction of psychological factors through quantitative numerical values and identify their priorities. In this study, the design and process are divided into three stages: Stage one is designing the questionnaire based on the DANP method and the model of mental health factors. In stage two, the questionnaire collects the practical experience of 33 hospital administrators by purposeful sampling method. Then, the degree of interaction of mental health factors is calculated by the DEMATEL method, and an influential network relationship map (INRM) is constructed. In stage three, the total influence matrix produced by the DEMATEL method is converted into a weight. The survey window was from May to June 2023, and the research design flow chart is shown in Figure 1.

Mental health factor model of work from home

Memon et al. (20) used the qualitative phenomenological design method to explore home office employees' life experiences during COVID-19 and recruited 41 employees using snowball sampling. The study (20) followed the thematic analysis steps defined by Braun and Clarke (21) to form a mental health factor model for home offices. In this study, we analyzed five themes surrounding working from home, namely technical problems, work-related stress, non-work stress, communication problems, and motivation and productivity problems. Because this study focuses on hospital administrators, technical issues, we excluded motivation and production and focused on work-related stresses. Therefore, non-work stresses and communication issues are excluded. In our study, we selected the appropriate dimensions and criteria for mental health factors, as shown in Table 1.

The DEMATEL and DANP methods

The DEMATEL method is a system structure analysis that analyzes the complex social network structure of problems in the real world using the practical experience of a group of experts (22, 23). This method can analyze the interaction between subsystems in a system and visualize it using graph theory, to construct an influence network diagram (24–26). This diagram can help decision-makers focus on a few major influencing factors (27, 28). Subsequently, a novel method has been developed that transforms the influence weights from the total influence relation results of the DEMATEL method through the

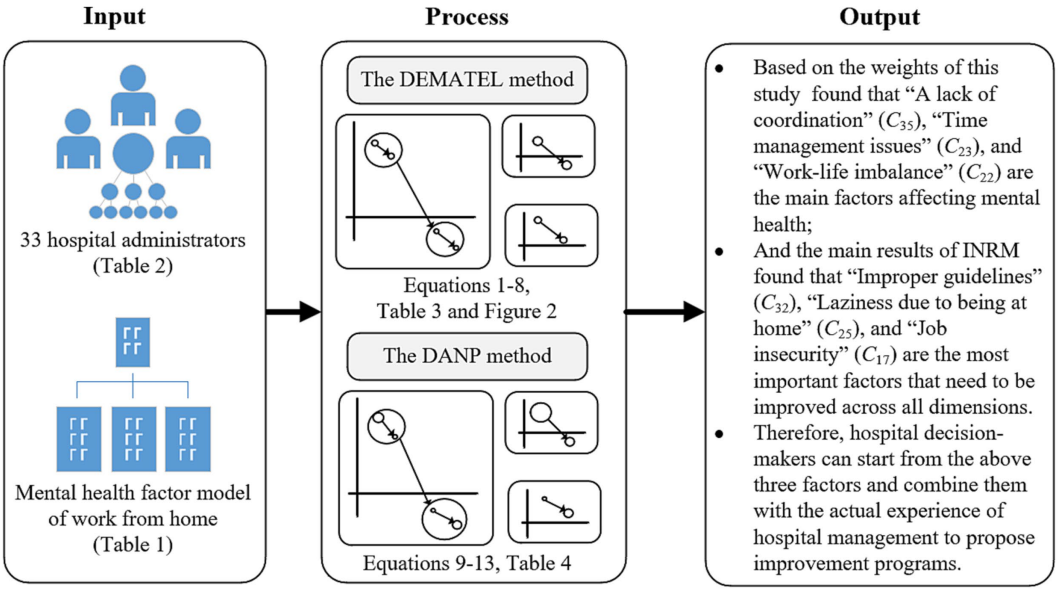

FIGURE 1
The research design flow chart.

TABLE 1 Mental health factor model of work from home.

Dimension	Criteria
Work-related stressors (C_1)	Workloads (C_{11})
	Work schedules (C_{12})
	Structural emptiness (C_{13})
	Unscheduled virtual meetings (C_{14})
	Weekend tasks (C_{15})
	High work expectations (C_{16})
	Job insecurity (C_{17})
Non-work stressors (C_2)	Distraction (C_{21})
	Work–life imbalances (C_{22})
	Time management issues (C_{23})
	Domestic issues (or children's presence at home) (C_{24})
	Laziness due to being at home (C_{25})
	An inconsistent sleep schedule (C_{26})
Communication issues (C_3)	Lack of social interaction (C_{31})
	Improper guidelines (C_{32})
	No feedback exchange (C_{33})
	No proper collaboration with superiors and peers (C_{34})
	A lack of coordination (C_{35})

principle and characteristics of the ANP method. This method is also called DEMATEL-based ANP or DANP. This method has been applied to policy management (29), food safety risk management (30), and online shopping (24). The detailed calculation steps of this method are outlined in previous related research (31–33). The mathematical steps and calculation equations of this method are as follows:

Step 1: Based on the mental health factor model of working from home, all experts quantify the mutual influence among all factors (That is, the degree of influence of factor i on factor j, and we ask the same thing again in reverse), then the factors must pass a set of quantitative scale of influence relationships (i.e., 0: *no influence* to 4 *extremely high influence*). The experience matrix $\boldsymbol{E} = \left[e_{ij}\right]_{n\times n}$ of each expert can be constructed, and the matrix $\boldsymbol{A} = \left[a_{ij}\right]_{n\times n}$ representing respondents can be obtained by the averaging method. See Equation 1.

$$\boldsymbol{A} = \begin{bmatrix} a_{11} & \cdots & a_{1j} & \cdots & a_{1n} \\ \vdots & \ddots & \vdots & \ddots & \vdots \\ a_{i1} & \cdots & a_{ij} & \cdots & a_{in} \\ \vdots & \ddots & \vdots & \ddots & \vdots \\ a_{n1} & \cdots & a_{nj} & \cdots & a_{nn} \end{bmatrix}_{n\times n} = \left[\frac{\left(\sum_{\varepsilon=1}^{\vartheta} e_{ij}^{\varepsilon}\right)}{\vartheta}\right]_{n\times n} \quad (1)$$

Step 2: Set the influence range boundary and convert the influence relationship degree to 0–1, as shown in Equations 2, 3.

$$\delta = \max\left\{\max\sum_{j=1}^{n} a_{ij}, \max\sum_{i=1}^{n} a_{ij}\right\} \quad (2)$$

$$\boldsymbol{D} = \frac{\boldsymbol{A}}{\delta} \quad (3)$$

Step 3: Calculate the total degree of mutual influence between factors, and finally, produce the total influence relationship matrix $\boldsymbol{T}$, as shown in Equation 4.

$$\boldsymbol{T} = \boldsymbol{D} + \boldsymbol{D}^2 + \ldots + \boldsymbol{D}^{\psi} = \boldsymbol{D}(\mathbf{I} - \boldsymbol{D})^{-1}, \text{when} \lim_{\psi\to\infty} \boldsymbol{D}^{\psi} = [0]_{n\times n} \quad (4)$$

Step 4: Derive the relevant analysis indexes of all factors, namely, "Give influence u_i," "Receive influence r_i," centrality, "Influence center, $u_i + r_i$" and "Influence cause or effect $u_i - r_i$" as shown in Equations 5–8.

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$$u_i = (u_1, u_2, \ldots, u_n) = \left[\sum_{j=1}^{n} t_{ij}\right]_{n\times 1} \tag{5}$$

$$r_i = (r_1, r_2, \ldots, r_n) = (t_j)'_{1\times n} = \left[\sum_{i=1}^{n} t_{ij}\right]'_{1\times n} \tag{6}$$

$$u_i + r_i \tag{7}$$

$$u_i - r_i \tag{8}$$

"Give influence u_i" and "Receive influence r_i" represent the influence of factors and the affected values, respectively. When these two indices are added, they represent the influence intensity of the factor in the whole system, while subtracting them indicates the influence nature of this factor in the system, that is, cause or effect. The former is called "Influence center $u_i + r_i$," and the latter is called "Influence cause or effect $u_i - r_i$."

Step 5: The boundary is established based on the total influence relation matrix $\boldsymbol{T}$ and converted into 0–1. The unweighted super matrix ω_C is derived, as shown in Equation 9.

$$\omega_C = (\boldsymbol{T}_C^{\alpha})' = \begin{array}{c} \begin{matrix} D_1 & \cdots & D_o & \cdots & D_m \end{matrix} \\ \begin{matrix} c_{11}\cdots c_{1m_1} & c_{o1}\cdots c_{om_o} & c_{m1}\cdots c_{mm_m} \end{matrix} \\ \begin{matrix} D_1 \\ \vdots \\ D_i \\ \vdots \\ D_m \end{matrix} \begin{matrix} c_{11} \\ \vdots \\ c_{1m_1} \\ c_{i1} \\ \vdots \\ c_{im_i} \\ c_{m1} \\ \vdots \\ c_{mm_m} \end{matrix} \begin{bmatrix} \boldsymbol{T}_{\alpha C}^{11} & \cdots & \boldsymbol{T}_{\alpha C}^{1o} & \cdots & \boldsymbol{T}_{\alpha C}^{1m} \\ \vdots & & \vdots & & \vdots \\ \boldsymbol{T}_{\alpha C}^{i1} & \cdots & \boldsymbol{T}_{\alpha C}^{io} & \cdots & \boldsymbol{T}_{\alpha C}^{im} \\ \vdots & & \vdots & & \vdots \\ \boldsymbol{T}_{\alpha C}^{m1} & \cdots & \boldsymbol{T}_{\alpha C}^{mo} & \cdots & \boldsymbol{T}_{\alpha C}^{mm} \end{bmatrix}_{n\times n | m<n, \sum_{o=1}^{m} m_o = n} \end{array} \tag{9}$$

Equation 10 shows the action of normalizing the total influence relation matrix (i.e., the values in the matrix are all between 0 and 1).

$$\boldsymbol{T}_{\alpha C}^{11} = \begin{array}{c} \begin{matrix} c_{11} & \cdots & c_{1o} & \cdots & c_{1m_1} \end{matrix} \\ \begin{matrix} c_{11} \\ \vdots \\ c_{1i} \\ \vdots \\ c_{1m_1} \end{matrix} \begin{bmatrix} t_{C11}^{11}/c_1^{11} & \cdots & t_{C1o}^{11}/c_1^{11} & \cdots & t_{C1m_1}^{11}/c_1^{11} \\ \vdots & & \vdots & & \vdots \\ t_{Ci1}^{11}/c_i^{11} & \cdots & t_{Cio}^{11}/c_i^{11} & \cdots & t_{Cim_1}^{11}/c_i^{11} \\ \vdots & & \vdots & & \vdots \\ t_{Cm_1 1}^{11}/c_{m_1}^{11} & \cdots & t_{Cm_1 j}^{11}/c_{m_1}^{11} & \cdots & t_{Cm_1 m_1}^{11}/c_{m_1}^{11} \end{bmatrix} \end{array} \tag{10}$$

$$= \begin{array}{c} \begin{matrix} c_{11} & \cdots & c_{1o} & \cdots & c_{1m_1} \end{matrix} \\ \begin{matrix} c_{11} \\ \vdots \\ c_{1i} \\ \vdots \\ c_{1m_1} \end{matrix} \begin{bmatrix} t_{\alpha C11}^{11} & \cdots & t_{\alpha C1o}^{11} & \cdots & t_{\alpha C1m_1}^{11} \\ \vdots & & \vdots & & \vdots \\ t_{\alpha Ci1}^{11} & \cdots & t_{\alpha Cio}^{11} & \cdots & t_{\alpha Cim_1}^{11} \\ \vdots & & \vdots & & \vdots \\ t_{\alpha Cm_1 1}^{11} & \cdots & t_{\alpha Cm_1 o}^{11} & \cdots & t_{\alpha Cm_1 m_1}^{11} \end{bmatrix}_{m_1\times m_1} \end{array}$$

Step 6: Transforming the unweighted super matrix into the weighted super matrix, i.e., the unweighted super matrix at the criterion level is adjusted by the conversion parameters at the dimension level, as shown in Equations 11, 12.

$$q^D = (\boldsymbol{T}_D^{\alpha})' = \begin{bmatrix} t_{11}^{D_{11}}/d_1 & \cdots & t_{1o}^{D_{1o}}/d_1 & \cdots & t_{1m}^{D_{1m}}/d_1 \\ \vdots & & \vdots & & \vdots \\ t_{i1}^{D_{i1}}/d_i & \cdots & t_{io}^{D_{io}}/d_i & \cdots & t_{im}^{D_{im}}/d_i \\ \vdots & & \vdots & & \vdots \\ t_{m1}^{D_{m1}}/d_m & \cdots & t_{mj}^{D_{mo}}/d_m & \cdots & t_{mm}^{D_{mm}}/d_m \end{bmatrix} = \begin{bmatrix} t_{11}^{\alpha_{11}} & \cdots & t_{1j}^{\alpha_{1o}} & \cdots & t_{1m}^{\alpha_{1m}} \\ \vdots & & \vdots & & \vdots \\ t_{i1}^{\alpha_{i1}} & \cdots & t_{io}^{\alpha_{io}} & \cdots & t_{im}^{\alpha_{im}} \\ \vdots & & \vdots & & \vdots \\ t_{m1}^{\alpha_{m1}} & \cdots & t_{mj}^{\alpha_{mo}} & \cdots & t_{mm}^{\alpha_{mm}} \end{bmatrix}_{m\times m} \tag{11}$$

$$\varpi = q^D \times \omega_C = \begin{bmatrix} t_{11}^{\alpha_{11}} \times \omega_C^{11} & \cdots & t_{i1}^{\alpha_{i1}} \times \omega^{i1} & \cdots & t_{m1}^{\alpha_{m1}} \times \omega^{m1} \\ \vdots & & \vdots & & \vdots \\ t_{1o}^{\alpha_{i1}} \times \omega^{1o} & \cdots & t_{io}^{\alpha_{io}} \times \omega^{io} & \cdots & t_{mo}^{\alpha_{mo}} \times \omega^{mo} \\ \vdots & & \vdots & & \vdots \\ t_{1m}^{\alpha_{m1}} \times \omega^{1n} & \cdots & t_{im}^{\alpha_{im}} \times \omega^{in} & \cdots & t_{mm}^{\alpha_{mm}} \times \omega^{mm} \end{bmatrix}_{n\times n | m<n, \sum_{o=1}^{m} m_o = n} \tag{12}$$

Step 6: The convergence process of the weighted super matrix through Markov chain calculation always reaches a steady state; that is, the influence weight of each factor is obtained, as shown in Equation 13.

$$\varpi^* = \lim_{\rho\to\infty} (\varpi)^{\rho} \tag{13}$$

Ethics approval

This study was approved by the Ethics Committee of Taizhou Central Hospital (Taizhou University Hospital) (Grant No. 2023L-05-07), it was conducted following the ethical guidelines described in the Declaration of Helsinki. The purpose was explained in detail to the experts before the investigation, and their consent was obtained during the investigation. Participants could terminate or withdraw from the study at any time during the study period.

Data collection and participants

The questionnaire is based on the characteristics of the DEMATEL method. At the same time, to increase the validity and reliability of data collection, the investigators adopted the purposive survey

TABLE 2 Demographic characteristics of 33 hospital administrators.

Characteristics	Value (%)
Sex	
Male	18 (55%)
Female	15 (45%)
Age	
<30	5 (15%)
30–39	19 (58%)
≥40	9 (27%)
Education	
Bachelor	21 (64%)
Master or above	12 (36%)
Years of service	
Under 10 years	13 (39%)
10–15	9 (27%)
15 and above	11 (34%)
Professional title	
Technologist-in-charge	3 (9%)
Supervisor nurse	3 (9%)
Senior technologist	2 (6%)
Senior nurse	5 (15%)
Senior engineer	1 (3%)
Senior doctor	1 (3%)
Senior accountant	2 (6%)
Researcher	1 (3%)
Registered nurse	2 (6%)
Librarian	1 (3%)
Engineer	1 (3%)
Economic engineer	1 (3%)
Doctor	3 (9%)
Chief nurse	1 (3%)
Associate professor	1 (3%)
Associate chief physician	2 (6%)
Accountant	3 (9%)
Experience in working from home	
Yes	33 (100%)
No	0 (0%)
Working hours from home	
<2 weeks	14 (42%)
2–3 weeks	10 (30%)
3–4 weeks	4 (12%)
≥4 weeks	5 (16%)

method and explained the purpose and significance of this study in person. Respondents agreed to participate in the study and fill out the questionnaire. Mental health survey data were collected from 33 hospital administrators who worked from home during the COVID-19 epidemic. The consensus gap of experts on this data result is 0.0121. In other words, the confidence level reaches 98.79% (i.e., the consensus gap is 1.12%). The survey was conducted from May to June 2023.

Results

Data presentation

In this questionnaire survey, there was little difference between men and women (55% men and 45% women), and their ages were mainly over 30 years ($n = 28$, 85%); most had a university education ($n = 21$, 64%), and most had worked for 10 years or more ($n = 20$, 61%). Furthermore, all respondents had practical experience of working from home and 58% worked from home for 2 weeks or more during the epidemic. The backgrounds of all respondents are shown in Table 2.

Network relation map

The relationship between psychological factors of working at home in 33 respondents during the COVID-19 epidemic can be analyzed by "Influence center $u_i + r_i$" and "Influence cause or effect $u_i - r_i$."

From the perspective of the "Influence center $u_i + r_i$," "Non-work stressors" (C_2) is the center of gravity for all mental health factors, and it has the highest interplay correlation compared to the other two dimension levels. Additionally, "Workloads" (C_{11}), "Work schedules" (C_{12}), and "Distraction" (C_{21}) were clearly the top three highest correlations of interactions with all factors compared to other mental health factors.

From the perspective of the "Influence cause or effect $u_i - r_i$," in the dimension level, the "Work-related stressors" (C_1) and "Non-work stressors" (C_2) are the effect groups; "Communication issues" (C_3) is the influence group. However, in the criteria level, "Workloads" (C_{11}), "Work schedules" (C_{12}), "Structural emptiness" (C_{13}), "Weekend tasks" (C_{15}), "Distraction" (C_{21}), "Work-life imbalance" (C_{22}), "Time management issues" (C_{23}), "Domestic issues (or children's presence at home)" (C_{24}), "Lack of social interaction" (C_{31}), "No proper collaboration with superiors and peers" (C_{34})" are the effect group; "Unscheduled virtual meetings" (C_{14}), "High work expectations" (C_{16}), "Laziness due to being at home" (C_{25}), "An inconsistent sleep schedule" (C_{26}), "Improper guidelines" (C_{32}), "No feedback exchange" (C_{33}), and "A lack of coordination" (C_{35}) are the cause group. Table 3 shows the results of the impacts of all factors and further shows the structure of the interrelationships between all factors by means of Figure 2, i.e., the influential network relation map (INRM).

Influence weight analysis

The influence weight represents the degree of influence of a factor on mental health in work from home. The higher the value, the more attention should be paid to this factor. For the dimensions, "Non-work stressors" (C_2) has the highest influence weight, followed by "Communication issues" (C_3) and "Work-related stressors" (C_1), from the local perspective. Furthermore, the highest weights in each

TABLE 3 The influential network structure of mental health factors.

Factors	Give influence	Receive influence	Influence center	Influence cause and effect
C_1	0.532	0.539	1.070	−0.007
C_{11}	5.043	5.197	10.240	−0.155
C_{12}	4.770	5.468	10.238	−0.699
C_{13}	4.241	4.248	8.489	−0.007
C_{14}	4.581	4.345	8.925	0.236
C_{15}	4.584	4.596	9.180	−0.013
C_{16}	4.745	4.735	9.480	0.010
C_{17}	4.813	4.427	9.240	0.386
C_2	0.748	0.751	1.498	−0.003
C_{21}	4.553	4.611	9.164	−0.057
C_{22}	4.965	5.056	10.021	−0.091
C_{23}	4.834	5.064	9.897	−0.230
C_{24}	4.316	4.564	8.880	−0.248
C_{25}	4.334	3.821	8.156	0.513
C_{26}	4.118	4.102	8.220	0.016
C_3	0.699	0.689	1.388	0.010
C_{31}	3.794	3.997	7.791	−0.202
C_{32}	4.555	4.070	8.625	0.484
C_{33}	4.218	4.158	8.375	0.060
C_{34}	4.109	4.195	8.304	−0.085
C_{35}	4.451	4.367	8.818	0.083

dimension are "Time management issues" (C_{23}), "A lack of coordination" (C_{35}), and "Work schedules" (C_{12}), which represent the local weight perspective. Finally, "A lack of coordination" (C_{35}), "Time management issues" (C_{23}), and "Work-life imbalance" (C_{22}) are the top three criteria in terms of the global perspective. The influence weight results for all the factors are listed in Table 4.

Discussion

Interpretation of findings based on the influence weight

In this study, "A lack of coordination" (C_{35}), "Time management issues" (C_{23}), and "Work-life imbalance" (C_{22}) are the key influencing factors. In the "Communication issues" (C_3) dimension, "A lack of coordination" (C_{35}) is a key influencing factor. A study of four Japanese manufacturing companies found differences in intra-company productivity between those who worked from home and those who did not during the COVID-19 pandemic. The results show that poor remote working environments, communication, and coordination are the main reasons for the decline in productivity (34). Poor communication in the workplace and with customers had significant negative effects. Face-to-face communication can effectively reduce the negative effects of uncoordinated communication. Amano et al. (35) found that during the COVID-19 pandemic, close communication between employees working at home and top leaders was a key factor affecting employee engagement. Another systematic review found that most studies showed that people who transition to work from home for the first time are most likely to be less productive than normal (36). Furthermore, one study showed that administrative staff who work remotely are worried about the lack of organizational communication and teamwork, which will affect their current work (37). In summary, a lack of communication and coordination may lead to mental health problems while working from home and may even aggravate the generation of negative emotions, resulting in a further reduction in productivity.

In the "Non-work stressors" (C_2) dimension, "Time management issues" (C_{23}) is an influencing factor. One study showed (38) that during periods when employees perform some or all of their job responsibilities at home, the time spent on childcare, housework, family dining, and preparation also increases significantly. Furthermore, the study found that even on the premise that the processing time of family affairs increased greatly, the time spent in remote work also showed an increasing trend. Therefore, people who work remotely from home experience great pressure on time management and need to pay more attention to it. Hospital administrators working remotely experience negative mental health effects.

In the "Non-work stressors" (C_2) dimension, "Work-life imbalance" (C_{22}) is another key influencing factor. In a literature review on the impact of COVID-19 on telecommuting employees, employees who were forced to switch to telecommuting because of the pandemic faced work–family conflicts and work overload, which can generate greater stress, accelerate fatigue, and reduce telecommuting satisfaction and job performance (36). In addition, Chu, Chan (39) showed that it is very important for management to maintain a healthy work-life balance for employees who work from home to support their mental health and improve their work efficiency. Among the three stress relief methods studied, work-life balance is the only one that affects employees' mental health. Therefore, during remote work, hospital administrators experience conflicts between work and family, which may lead to negative emotions and affect their mental health.

Implications based on the INRM

"Influence center" and "Influence cause or effect" can show the structure of the network relationship between all factors, namely the INRM, as shown in Figure 2. The Figure shows that "Improper guidelines" (C_{32}), "Laziness due to being at home" (C_{25}), and "Job insecurity" (C_{17}) are the most important factors needing improvement in all dimensions. Therefore, hospital decision-makers can propose improvement schemes from the above three factors and combine them with their practical experience in hospital management.

During the COVID-19 pandemic, telecommuting is no longer a unique working mode but has become an effective supplement to the traditional working mode. Hospital administrators' main tasks include writing documents, data analysis, and communication. Incorrect guidelines will affect the performance of remote work, which will lead to poor work outcomes and indirectly cause psychological pressure.

This study concludes that addressing "Improper guidelines" (C_{32}) is an effective improvement factor, and improvement measures can

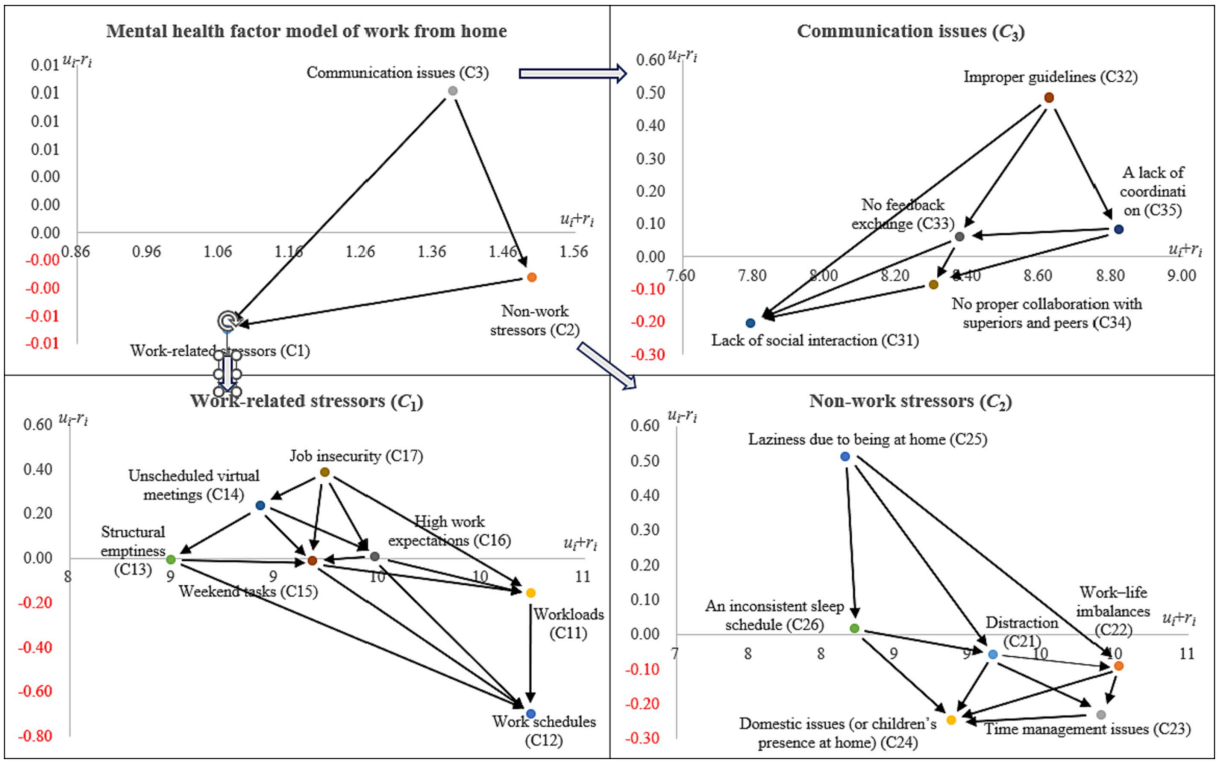

FIGURE 2
The influential network relation map (INRM).

be proposed from two levels of hospital managers and hospital administrative staff. At the management level, hospital leaders and department managers need to acknowledge that telecommuting has become an indispensable part of their daily work, and the advantages and disadvantages of telecommuting, including productivity, job performance, and mental health, must be fully considered.

The guidelines issued by leaders have a significant influence on the aforementioned telecommuting problems of managers and cannot provide work guidance according to the traditional working mode. Therefore, the guidelines for remote work at the leadership level, including work content, completion, quality requirements, and data collection, should be clear. Managers need to actively communicate fully with the administrative staff who work remotely, listen to the opinions of the work implementers, and adjust the contents of the guidelines to ensure they are practical and can be implemented remotely.

In addition, management styles can lead to inappropriate guidelines. Such leaders often have high authority, do not allow others to express their opinions about work, and require attention. When hospital administrators receive remote work instructions from managers, they should not blindly implement them and instead immediately confirm the work content, completion time, and work requirements with directly affiliated managers. Before starting work, the feasibility of the remote working mode should be analyzed, and opinions for managers' reference should be put forward to avoid improper guidance affecting remote work.

This study found that addressing "(C_{25})" laziness caused by being at home is another effective improvement factor. When administrators work in hospitals, the working environment includes constraints and supervision factors such as working hours and peer supervision, which can ensure work efficiency. When working remotely, the restrictions and supervision factors of the hospital working environment disappear, and laziness occurs at home, leading to lower work efficiency and longer working hours. Improvement strategies can be proposed based on these five perspectives.

In terms of personal ability, hospital administrators can improve their self-discipline and time management ability. Self-discipline is the primary factor that affects personal work efficiency. Some studies have shown that employees who think they are self-disciplined are more active, effective, and timely in time management than those who think they are not (36). Therefore, improving the quality of self-discipline and time management ability of hospital administrators can improve the efficiency of remote work and reduce negative emotions such as anxiety and uneasiness caused by laziness and lack of self-discipline.

At the hospital organization system level, hospitals should supervise the remote work of administrators and adopt flexible working hours. Supervision is an important management strategy for overcoming laziness. Hospitals can supervise the effects of telecommuting through regular work reports and inspections, urge telecommuting managers to begin work on time, avoid unreasonable time arrangements caused by laziness, objectively reduce the probability of laziness at home, and reduce the negative emotions and psychological states caused by laziness. Flexible working hours should be adopted. Anyone working remotely has their own unique and efficient working hours. By adopting a flexible working-hours mode, teleworkers can use their working hours efficiently to complete their work and objectively reduce their laziness.

TABLE 4 The influential weights of mental health factors.

Factors	Local weight	Rank	Global weight	Rank
C_1	0.271	3		
C_{11}	0.158	2	0.043	13
C_{12}	0.166	1	0.045	12
C_{13}	0.129	7	0.035	18
C_{14}	0.131	6	0.036	17
C_{15}	0.139	4	0.038	15
C_{16}	0.143	3	0.039	14
C_{17}	0.135	5	0.037	16
C_2	0.380	1		
C_{21}	0.169	3	0.064	8
C_{22}	0.186	2	0.071	3
C_{23}	0.186	1	0.071	2
C_{24}	0.168	4	0.064	9
C_{25}	0.141	6	0.053	11
C_{26}	0.151	5	0.057	10
C_3	0.349	2		
C_{31}	0.192	5	0.067	7
C_{32}	0.196	4	0.068	6
C_{33}	0.200	3	0.070	5
C_{34}	0.202	2	0.070	4
C_{35}	0.210	1	0.073	1

At the hospital logistics support and humanistic care levels, hospital administrators should control fatigue when working remotely. Some studies have shown that owing to factors such as home place, office conditions, and working hours, the probability of muscle soreness and eye fatigue in home telecommuting is higher than that in office places (38). From another perspective, fatigue and discomfort lead to increased fatigue in telecommuters, and some administrators may increase the likelihood of laziness. Therefore, hospitals should provide hardware support for telecommuters, such as ergonomic office chairs and proper lighting, which can effectively reduce fatigue while working from home. At the psychological level, such people can also feel the support of the organization, improve their motivation within the work, and reduce the possibility of laziness.

This study also found that addressing "Job insecurity" (C_{17}) and job instability are also effective improvement factors. Hospital administrators generally have clear job responsibilities and work plans, and telecommuting leads to significant changes in their work content, workload, and working hours. These aspects are unstable, and hospital administrators are prone to psychological pressures, such as anxiety and irritability. Therefore, improvement strategies are proposed for these three levels.

Regarding the work content level, additional tasks may be added during remote work, including newly added temporary work, to enable collaboration with colleagues to complete the work. During the COVID-19 pandemic, hospital administrators added considerable temporary work to data reporting and documentation and needed to provide work assistance to colleagues who were resting. The above work contents are all new tasks, and administrators who need to work remotely are particularly unfamiliar with the new process and work content, which causes tension and anxiety.

When managers arrange new tasks, they should plan and decompose the work content and arrange people with similar work content or relevant skills. In addition, we should pay attention to the problems encountered in the process of carrying out new work and help solve them promptly. Facing new work tasks, managers who work remotely adjust their psychological state over time, make work plans and support conditions, and report to them to obtain work guidance and support (39).

Regarding workload and working hours, this depends mainly on the task itself and organizational factors. Each task has its own work content and time-limit requirements, which directly determine the workload and working hours. Managers should consider the sum of the workload and working hours of each executive and try their best to achieve balance.

Organizational factors include organizational design and leadership style. For example, during the COVID-19 pandemic, the medical management department undertook most of the prevention and control management and data statistics, as well as much coordination work. The new workload and working hours increased significantly, but compared with other administrative departments, there was no obvious increase. In view of this phenomenon, breaking the traditional bureaucratic structure and implementing the project structure in some posts can effectively adjust the workload and working hour pressure of key departments.

In addition, a positive leadership style can elicit positive emotions from team members, making employees feel that their organization is taking care of them and that their work can develop positive emotional resources. In these cases, current and caring leadership styles represent an appropriate form of organizational support that can effectively reduce the psychological stress caused by bad emotions.

Limitations

This study had several limitations. First, the participants in this study were recruited through a purposeful sampling method, which may have led to sampling deviation. In addition, the results were limited to the investigation of the case hospital at that time and should not be inferred from subsequent time points or other hospitals. Finally, the method used in this study aimed to obtain the influence network structure and corresponding weights from the perspective of influence, which is different from the preference relationship weighting method (such as the analytic hierarchy process).

Conclusion

Based on the weights of this study and the main results of INRM found that "A lack of coordination" (C_{35}), "Time management issues" (C_{23}), and "Work-life imbalance" (C_{22}) are the main factors affecting mental health; and that "Improper guidelines" (C_{32}), "Laziness due to being at home" (C_{25}), and "Job insecurity" (C_{17}) are the most important factors that need to be improved across all dimensions. Therefore, hospital decision-makers can start from the above three factors and combine them with the actual experience of hospital management to

propose improvement programs. Also, scholars can further study the mental health factors of home-based workers from different perspectives, including different roles (i.e., teachers), different health factors (i.e., adding other factors), and analyzing different decision-making methods (i.e., from different decision-making perspectives). These are all future research directions that will help hospital decision-makers take early preventive measures for home office mental health problems in the face of potential major infectious diseases in the future.

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 humans were approved by the Ethics Committee of Taizhou Central Hospital (Taizhou University Hospital) (Grant no. 2023L-05-17) and was conducted in accordance with the ethical guidelines described in the Declaration of Helsinki. The purpose was explained in detail to the experts before the investigation, and their consent was obtained during the investigation. Participants could terminate or withdraw from the study at any time during the study. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

SS: Writing – original draft, Investigation. JZ: Writing – original draft, Formal analysis. WS: Data curation, Methodology, Writing – original draft. Y-CC: Methodology, Supervision, Writing – original draft, Writing – review & editing. CL: Formal analysis, Validation, Writing – original draft. HL: Project administration, Supervision, Writing – review & editing.

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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.

Publisher's note

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Factors associated with patients' healthcare-seeking behavior and related clinical outcomes under China's hierarchical healthcare delivery system

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Introduction: The hierarchical healthcare delivery system is an important measure to improve the allocation of medical resources and promote equitable distribution of basic medical and health services. It is one of the key factors in the success or failure of China's medical reform. This study aims to analyze the factors influencing patients' healthcare-seeking behaviors, including socioeconomic and clinical outcomes, under China's hierarchical healthcare delivery system, and to provide potential solutions.

Methods: Patients receiving outpatient treatment in the past 14days and inpatient care in the past 1year were investigated. The multivariate logistic regression was used to analyze the influencing factors of patient's medical treatment behavior selection, and to compare whether the clinical outcomes of primary medical institutions and grade A hospitals are the same.

Results: Nine thousand and ninety-eight person-times were included in the study. Of these, 4,538 patients were outpatients, 68.27% of patients were treated in primary medical institutions; 4,560 patients were hospitalized, 58.53% chose to be hospitalized in grade A hospitals. Provinces and cities, urban and rural areas, occupation, education level, medical insurance type, income, whether there are comorbid diseases, and doctors' medical behavior are the factors affecting the choice of medical treatment behavior. Patients who choose primary medical institutions and grade A hospitals have different control levels and control rate for the blood pressure, blood lipids, blood glucose.

Conclusion: Under the hierarchical diagnosis and treatment system, the patients' choice of hospital is mainly affected by their level of education, medical insurance types, and the inpatients are also affected by whether there are comorbid conditions. Clinical outcomes of choosing different levels of hospitals were different.

KEYWORDS

patients' healthcare-seeking behavior, outcomes, hierarchical healthcare delivery system, cardiovascular risk factors, primary healthcare

Introduction

The hierarchical healthcare delivery system is one of the effective measures for countries to improve the allocation of medical resources and improve the efficiency of diagnosis and treatment (1, 2). In 2009, the CPC Central Committee and the State Council issued the Opinions on Deepening the Reform of the Medical and Health Care System, which proposed the concept of hierarchical diagnosis and treatment for the first time (3). In 2015, the General Office of the State Council issued the Guiding Opinions on Promoting the Construction of a Graded Diagnosis and Treatment System, actively exploring the establishment of a hierarchical diagnosis and treatment system across the country (4). Since China does not force patients to diagnose and treat according to the level, the proportion of medical treatment at the primary healthcare is still not ideal (5). Data shows that the current treatment rate of primary hospitals varies from 40–80% (6, 7). It is difficult to be treated in grade-A hospitals (8), and many of them are chronic patients (9), while the primary hospitals have very few patients there. Therefore, it is important to understand the influencing factors of patients' choice of medical treatment behavior (10). Several studies have shown that (11–14) the main reason affecting patients' choice of medical institutions is the quality of medical care. Past studies have assumed that the quality of chronic diseases treatment is the same in different medical institutions, but this may not be the case. Therefore, comparing the clinical indicators in different levels of medical institutions may find the main reasons affecting patient selection. This can guide the government departments to formulate corresponding policies and measures according to the actual influencing factors, which will help to solve the problem of low treatment rate of primary medical institutions in the hierarchical diagnosis and treatment.

Materials and methods

The study adopted a two-stage stratified clustering design to ensure a representative and unbiased sample across various provinces, which is elaborated by the random selection of communities or villages within each selected province. Initially, one province is randomly selected from 7 geographical regions in China (Northeast, North, northwest, east, Central, South, and southwest). Beijing, Xinjiang Uygur Autonomous Region, Henan, Jilin, Guangdong, Yunnan, Jiangxi, and Zhejiang provinces were randomly selected for further sampling. All residents in selected communities (villages) were surveyed from June 1, 2014 to December 31, 2016. The patients with cardiovascular risk factors who are inpatient within 1 year and outpatient within 2 weeks were included in the study. A face-to-face structured interview were carried out by trained researchers using standard questionnaires and the physical examinations and laboratory tests were completed as well. In the morning, a fasting venous blood sample was taken for measurements of blood glucose, total cholesterol, low-density lipoprotein cholesterol (LDL-C) and triglycerides. The laboratory tests were carried out by Guangzhou Jinyu medical laboratory center Co., Ltd which was certified by the College of American Pathologists.

Definitions

Cardiovascular risk factors and related diseases definitions: Factors (15) used in the 10-year CVD (CVD) risk model (China) included: age, mean systolic blood pressure, fasting total cholesterol, HDL cholesterol, current smoking (yes/no), diabetes (yes/no), body mass index (BMI), waist circumference, geographic area (northern/southern China), urban/rural residents and family history of CVD (16). Smoking status was defined as a self-reported non-smoker, former smoker (1 year), or current smoker. Diabetes was defined as fasting blood glucose 7.0 mmol/L, antidiabetic medication, or previous postprandial blood glucose 11.1 mmol/L, HbA1c 7%, or a diagnosis of diabetes. Weight, height, and waist circumference were measured by trained researchers using standard methods. Beijing, Xinjiang Uygur Autonomous Region, Henan and Jilin Province are defined as northern China, while Guangdong, Yunnan, Jiangxi, Zhejiang and other provinces are defined as southern China (17). Family history of CVD was defined as a history of coronary heart disease or stroke in any of the participant's parents or immediate brother or sister. Hypertension was defined as measuring systolic blood pressure (SBP)140 mmHg, diastolic blood pressure (DBP) 90 mmHg (18), taking antihypertensive drugs, or self-reporting a previous diagnosis of hypertension. We defined SBP <130 mmHg and DBP< 80 mmHg as blood pressure control (19). Dyslipidemia was defined as total cholesterol 240 mg/dL, low-density lipoprotein cholesterol (LDL-C) 160 mg/dL, taking statins or other lipid-lowering drugs, or previously diagnosed as dyslipidemia. Cholesterol and blood glucose were measured in a central laboratory certified by the College of American Pathologists (Guangzhou Kimmer Testing Technology Co., Ltd).

Questionnaire design

The questionnaire was designed by the China Clinical Research Center for Cardiovascular Disease (20) based on the national health service survey, the epidemic survey, the influential published questionnaires, and the opinions of several experts. For questionnaire validation, we conducted a presurvey of 400 respondents in a community, verified the internal and external validity of the questionnaire and modified it to the questions found. The final

Abbreviations: CVD, cardiovascular disease; GAH, grade A hospital; HHDS, hierarchical healthcare delivery system; PH, primary healthcare; PHSB, patient healthcare seeking behavior.

questionnaire includes: basic personal information, disease-related information, quality of life, health service utilization, clinical results and patient self-assessment, etc. (21).

Theoretical framework

In our study, the analysis will be performed in the four dimensions of Andersen's model (22) with further exploration of the interactions among the various dimensional factors.

The Anderson Healthcare Services behavior utilization model was established in 1968 by Ronald Max Anderson (23). It was originally used to analyze the influencing factors of home health service utilization. In the current Andersen's model, after five modifications, patient medical care seeking behavior was influenced by four dimensions: situational characteristics, personal characteristics, medical behavior and outcome. There are interactions among these factors.

Variable

The study data included situational characteristics (province and city, urban and rural), personal characteristics [age, gender, marital status, education level, family income and medical coverage, combination with hypertension, diabetes, dyslipidemia, coronary heart disease, stroke/transient ischemic attack (TIA) and atrial fibrillation; lifestyle factors included smoking and alcohol consumption]. Medical behavior (e. g., whether blood pressure is measured, whether to explain the significance of blood pressure values, and whether patients with chronic diseases prescribe corresponding drugs). And results (patient evaluation of their health status, current blood pressure (systolic and diastolic blood pressure), blood glucose, lipids, waist circumference, BMI levels), and disease awareness rate, treatment rate, and control rate.

Quality control

Professor Du Xin, Professor Ma Changsheng and Professor Dong Jianzeng were responsible for designing the questionnaire and verifying the internal and external validity of the questionnaire. A group of regular personnel led by Guo Lizhu and Yang Xiaohui guide the national investigators to complete the questionnaire. Before the survey, the investigators were given unified training. For respondents with low education or illiterate, the investigator asked them questions and the questionnaire were filled out by the investigator. The study was approved by the ethics committee, and written informed consent was obtained from the respondents for each questionnaire. The questionnaire was answered on portable Android device (PAD), as a structured electronic questionnaire. The questionnaire is designed with reversed question verification the logical relationship, the numerical range is limited to avoid filling errors. If the questionnaire with contradictory answers, missed answers or errors cannot be submitted, and the errors are corrected on the spot. Effectiveness validation was performed after data collection by Du Jing, Wang Chi and Wu Huanqi. Xia Shijun made a preliminary analysis of part of the data.

Statistical analysis

Through uni-variate analysis, compares the differences between patients in four dimensions: background characteristics, individual characteristics, health behaviors and outcome indicators, further incorporates statistically significant factors into the model, and explores the independent factors influencing selection of patients' medical seeking behavior through multiple logistic regression analysis.

Categorical variables are shown as *n* (%), and continuous variables are shown as mean (SD). Continuous variables will be compared by using the unpaired t test, categorical variables by using the χ^2 test. Using the SAS PROC SURVEYLOGISTIC procedure, with adjustment for sociodemographic and clinically relevant covariates, including age, sex, area of residence (urban versus rural), region, education level, household income, insurance status, and occupations, to assess the association between these factors and health seeking behaviors.

The clinical outcomes of blood pressure, blood glucose, and blood lipids in patients treated in primary healthcare and Grade-A hospitals were further compared.

Results

The sampled community population is 47,841 people among which 24,344 people had risk factors for cardiovascular disease, 9,098 person-times had medical experience, 4,538 people visited outpatient clinics within 2 weeks, and 4,560 people had been hospitalized within 1 year. The statistics of urban and rural population according to outpatient visits and inpatient treatment and the number of people selected in this study by province are shown in Table 1.

Table 2, by INPATIENTS choice of primary healthcare (PH) or grade A hospitals (GAH), the baseline characteristics, medical behaviors experienced, and some healthcare outcomes of the participants were listed. Among the 4,538 participants, 1,441 (31.75%) people chose the grade A hospital. Compared with patients who chose primary healthcare facilities, patients living in urban areas (PH: 43.79%; GAH: 29.29%), lives in northern China (PH: 37.85%; GAH: 44.14%), people engaged in non-manual labor (PH: 20.34%; GAH: 34.56%), high school education level or above (PH: 16.05%; GAH: 28.31%), the upper middle class of household income (50,000–70,000 yuan) (PH: 13.53%; GAH: 16.59%), with urban medical insurance (PH: 67.71%; GAH: 88.48%), combined with cardiovascular disease (PH: 3.07%; GAH: 6.04%), married person (PH: 83.76%; GAH: 85.01%), Han (PH: 96.22%; GAH: 97.29%) prefer to choose the grade A hospital. There were no significant differences in the gender or age of patients. In the course of medical behavior, patients who chose to visit primary healthcare received a higher proportion of blood pressure measurements during the visit (PH: 57.87%; GAH: 50.37%), the results of blood pressure measurement interpreted by the doctor (PH: 97.73%; GAH: 96.61%), but fewer correctly prescribed drugs were given (PH: 62.97%; GAH: 69.97%). A larger proportion of patients visited primary healthcare considered themselves to be healthy (PH: 94.30%; GAH: 92.62%), rate of blood pressure control is lower (PH: 64.13%; GAH: 71.20%).

Table 3, by INPATIENTS 'choice of primary healthcare or grade A hospitals, the baseline characteristics, medical behaviors experienced, and some healthcare outcomes of the participants were listed. Among the 4,560 participants, 2,669 (58.53%) people

TABLE 1 The number of participants in the national survey is grouped by urban/rural areas and provinces.

	N(%)	Primary healthcare		Grade-A hospital		Primary healthcare		Grade-A hospital	
Cities									
Rural	3,058	1,110	(35.84)	112	(7.77)	1,220	(64.52)	616	(23.08)
Urban	6,040	1987	(64.16)	1,329	(92.23)	671	(35.48)	2053	(76.92)
Provinces									
Beijing	1,185	444	(14.34)	421	(29.22)	48	(2.54)	272	(10.19)
Guangdong	1721	774	(24.99)	421	(29.22)	164	(8.67)	362	(13.56)
Henan	1,286	339	(10.95)	117	(8.12)	292	(15.44)	538	(20.16)
Jilin	1,088	221	(7.14)	70	(4.86)	352	(18.61)	445	(16.67)
Jiangxi	424	98	(3.16)	79	(5.48)	41	(2.17)	206	(7.72)
Xinjiang	805	168	(5.42)	28	(1.94)	375	(19.83)	234	(8.77)
Yunnan	1,556	427	(13.79)	117	(8.12)	552	(29.19)	460	(17.23)
Zhejiang	1,033	626	(20.21)	188	(13.05)	67	(3.54)	152	(5.7)

TABLE 2 Characteristics of influencing factors (number and percentage) of INPATIENTS at cardiovascular risk factors when choosing different medical healthcare institution for treatment.

	Primary healthcare *n* = 3,097	Grade A hospital *n* = 1,441	*p*
Environment			
District (rural), %	1,110(35.84)	112(29.29)	<0.0001
Province (north), %	1,172(37.85)	636(44.14)	<0.0001
Population characteristics			
Gender (man), %	1,054(34.03)	468(32.48)	0.3015
Age (<65 years), %	1977(63.84)	922(63.98)	0.7952
Ethnic(han), %	2,980(96.22)	1,402(97.29)	0.2797
Marital status (married), %	2,594(83.79)	1,225(85.01)	0.0185
Occupation (mental), %	630(20.34)	498(34.56)	<0.0011
Education (below junior high school), %	2,156(66.66)	753(57.01)	<0.0001
Income (5–70,000), %	419(13.54)	239(16.61)	<0.0001
Health insurance (urban health insurance), %	2097(67.71)	1,275(88.48)	<0.0001
Disease (CHD), %	95(3.07)	87(6.04)	0.0002
Health behavior			
Measure BP	1,659(53.57)	682(47.33)	<0.0001
Inform the patient of HT	2,802(97.73)	1,308(96.60)	0.1878
Treated rate of HT, %	1,170(62.97)	557(69.97)	0.0005
Outcome			
Self evaluation (healthy), %	2,911(94.30)	1,331(92.62)	<0.0001
HT awareness rate	1,387(74.65)	608(76.38)	0.1014
Hypertension control rate	747(40.20)	381(47.86)	0.0003

chose the grade A hospital. Compared with patients who chose primary healthcare facilities, patients living in urban areas (PH: 35.48%; GAH: 76.92%), lives in southern China (PH: 43.58%; GAH: 44.21%), male (PH: 41.85%; GAH: 36.28%), people engaged in non-manual labor (PH: 11.42%; GAH: 24.73%), high school education level or above (PH: 15.66%; GAH: 34.03%), the upper middle class of household income (3–70,000 yuan) (PH: 25.30%; GAH: 35.92%), with medical insurance and which is not the new rural cooperative medical insurance (PH: 38.66%; GAH: 72.35%), combined with coronary heart disease (CHD) (PH: 6.28%; GAH: 11.13%), Han (PH: 88.74%; GAH: 95.28%) prefer to choose the grade A hospital. There were no significant differences in the

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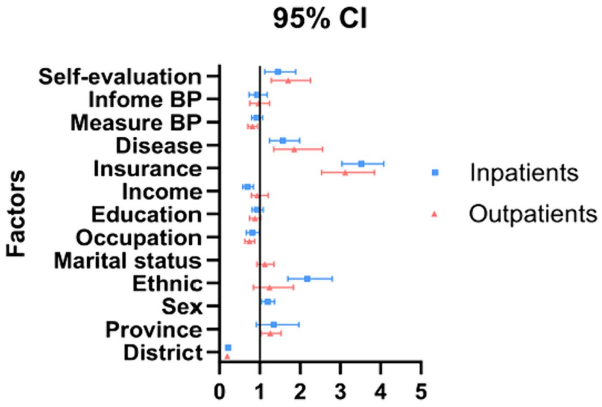
TABLE 3 Characteristics of influencing factors (number and percentage) of INPATIENTS at cardiovascular risk factors when choosing different medical healthcare institution for treatment.

	Primary healthcare $n = 1891$	Grade A hospital $n = 2,669$	p
Environment			
District (rural), %	1,220(64.52)	616(23.08)	<0.0001
Province (north), %	1,067(56.42)	1,489(55.79)	<0.0001
Population characteristics			
Gender (man), %	686(36.28)	1,117(41.85)	0.0001
Age (<65 years), %	1,061(56.11)	1,451(54.36)	0.1880
Ethnic(han), %	1,678(88.74)	2,543(95.28)	<0.0001
Marital status (married), %	1,566(82.86)	2,225(83.40)	0.1707
Occupation (mental), %	216(11.42)	660(24.73)	0.0526
Education (below junior high school), %	1,594(84.34)	1760(65.97)	<0.0001
Income (3–70,000), %	478(25.30)	958(35.92)	<0.0001
Health insurance (urban health insurance), %	731(38.66)	1931(72.35)	<0.0001
Disease (CHD), %	129(6.82)	297(11.13)	0.002
Health behavior			
Measure BP	726(40.31)	1,097(42.27)	0.2019
Inform the patient of HT	1,693(97.92)	2,423(97.16)	<0.0001
Treated rate of HT, %	606(49.55)	1,059(62.40)	<0.0001
Outcome			
Self-evaluation (healthy), %	1751(93.48)	2,449(92.25)	0.0121
HT awareness rate	867(70.89)	1,274(75.07)	0.0066
Hypertension control rate	402(32.87)	622(36.65)	0.0005



variable	Outpatients OR	95% CI		Inpatients OR	95% CI	
District	0.189	0.144	0.248	0.218	0.177	0.269
Province	1.259	1.037	1.529	1.345	0.917	1.972
Sex				1.195	1.046	1.364
Ethnic	1.241	0.839	1.835	2.179	1.698	2.795
Marital status	1.122	0.931	1.353			
Occupation	0.741	0.629	0.872	0.814	0.667	0.994
Education	0.878	0.748	1.029	0.936	0.807	1.086
Income	0.942	0.792	1.21	0.696	0.576	0.84
Insurance	3.12	2.532	3.846	3.522	3.041	4.08
Disease	1.856	1.347	2.558	1.572	1.242	1.989
Measure BP	0.818	0.705	0.948	0.924	0.797	1.072
Infome BP	0.966	0.753	1.238	0.934	0.737	1.183
Self-evaluation	1.701	1.282	2.257	1.456	1.12	1.893

FIGURE 1
Analysis of the influencing factors for patients at risk of cardiovascular disease when choosing different medical institutions for treatments.

marital status or age of patients. In the course of medical behavior, INPATIENTS who chose to visit grade A hospitals received result interpretation of their blood pressure measurements less than primary healthcare during the hospitalization (PH: 97.92%; GAH: 97.16%), patient in grade A hospital more correctly prescribed drugs were given (PH: 49.55%; GAH: 62.40%). There is no difference in the proportion of patients given by doctors to measure blood pressure in primary healthcare and grade A hospitals. Patients attending grade A hospitals perceived their health to be worse (PH: 6.52%; GAH: 7.75%), more people know they have high blood pressure (PH: 75.07%; GAH: 70.89%), higher blood pressure control rate (P: 32.87%; GAH: 36.65%).

Figure 1 shows the results of binary logistic regression analysis of factors affecting patients' medical behavior choices. Whether

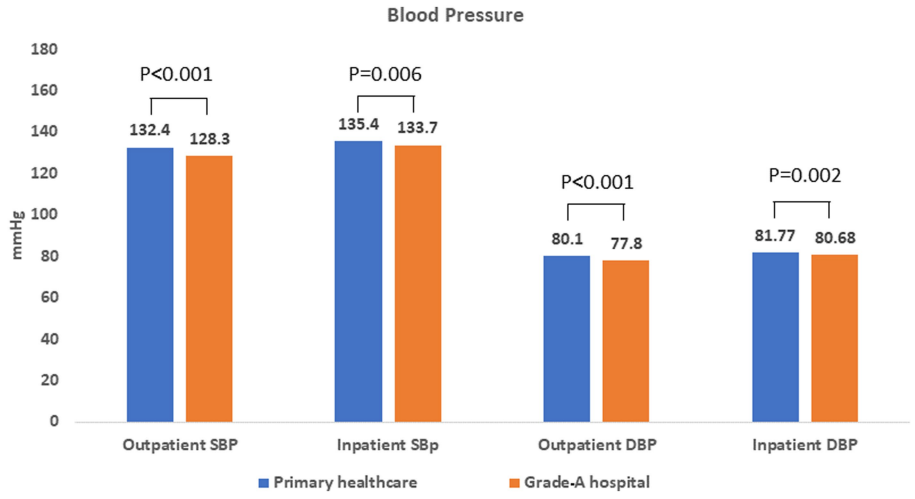


FIGURE 2
Blood pressure of patient visits to different levels of medical institutions.

INPATIENT OR OUTPATIENT, urban and rural areas, their own health evaluation, combined diseases, their own occupation and type of medical insurance are the factors that influence patients' choice of medical treatment. Their resident city and whether the doctor measure the patient's blood pressure at the time of the visit are the factors that affect the patient's OUTPATIENT visit. The amount of income, ethnic and gender affects the choice of INPATIENT hospitalization medical behavior.

Figure 2 shows the current blood pressure levels of patients who chose primary healthcare or grade A hospitals being OUTPATIENT and INPATIENT, respectively. It can be seen that patients at different levels of medical institutions, the patients' blood pressure difference, whether systolic or diastolic, whether OUTPATIENT or INPATIENT. Patients have better blood pressure levels after treatment in grade A hospitals (SBP 128.3 ± 18.3 mmHg, DBP 77.8 ± 10.2 mmHg), among which OUTPATIENTS who visited grade A hospitals have the best blood pressure control, and the average value reaches the target blood pressure (≤130/80 mmHg) (M).

Cohen's kappa was employed to ensure consistency in hypertension diagnosis among patients, yielding high agreement rates (0.9793 for outpatients and 1 for inpatients).

Figure 3 shows the current clinical outcomes of patients who chose primary healthcare and grade A hospitals being OUTPATIENT AND INPATIENT respectively: blood glucose, total cholesterol, LDL cholesterol. It can be seen that INPATIENTS at different levels of hospitals have different levels of blood glucose, total cholesterol, and LDL cholesterol levels. The patients have better clinic outcome after treatment in grade A hospitals ($TC_{OUTPATIENT}$ 5.19 ± 1.00 mmol/l, $TC_{INPATIENT}$ 5.07 ± 1.05 mmol/l; $LDL_{INPATIENT}$ 3.03 ± 0.87 mmol/l) and lower levels of total cholesterol and LDL cholesterol after outpatient treatment in grade A hospitals ($LDL_{OUTPATIENT}$ 3.13 ± 0.85 mmol/l). The former is statistically different. Whether OUTPATIENT AND INPATIENT, current blood glucose after treatment at different levels of medical healthcare is up to standard.

Discussion

In this study, the rates of outpatient and inpatient visits primary hospitals with cardiovascular risk factors were 68.27 and 41.47% respectively, which were lower than the policy target proposed by the Chinese government in 2015 (70%). We found that the main factors influencing the choice of patient medical treatment behavior are occupation, education, reimbursement method, income, and whether the patient had a comorbid illness. Secondly, marital status, living habits and body shape affect the choice of OUTPATIENT medical treatment. Gender and ethnic group influence the INPATIENTS' choice of hospitalization (24).

Hierarchical diagnosis and treatment refer to the classification according to the severity of diseases and the level of difficulty of treatment, the medical institutions of different levels undertake the treatment of different diseases (25–27). Mix all the diseases together to discuss the effect of hierarchical diagnosis and treatment is not effective nor conducive to identify the potential reasons affecting patients' behavioral choice.

This study was sampled in the national wide communities and focused on patients with CVD risk factors, the vast majority of whom were with hypertension. This is conducive to the analysis of the medical behavior of cardiovascular patients who should go to the primary hospital for diagnosis and treatment. As is known to all, cardiovascular disease is the first burden disease in China which has the large number of patients and was the heavy medical burden (27–29). Patients with chronic cardiovascular diseases such as hypertension did not act in according to the recommended program (primary medical healthcare treatment) is one of the main reasons leading to the failure of graded diagnosis and treatment (30, 31).

Anderson's theoretical framework (32) shows that patient medical treatment behavior choice is influenced by four dimensions: environmental factors, personal factors, medical behavior and medical outcome. For such significant number of hypertension patients, on the facts of the extensive countrywide publication, the diagnosis and treatment costs are not high, and almost all medical institutions can

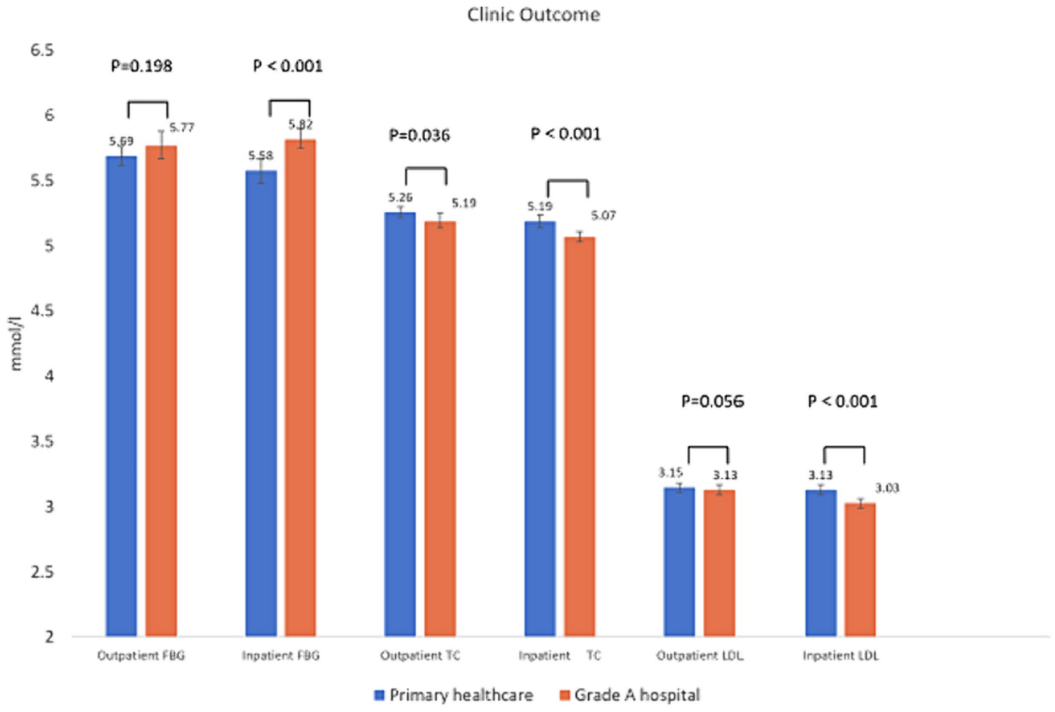


FIGURE 3
Blood glucose and lipid status of outpatient visits to different levels of medical institutions.

provide treatment, why there are still many patients choose the grade-A hospital? It is worth our thinking.

Hypertension is a common disease, a high incidence of disease, when we do the evaluation of health service utilization, we often assume that the clinical effect of treating hypertension in any medical institution to treat hypertension is the same, but is it really the case? (33, 34).

In addition to the questionnaire, in this study we also measured the clinical indicators of the patients, which enabled us to evaluate the control effect of the disease. Through the data, we found that the blood pressure and lipid levels of patients in primary medical institutions were higher than those of patients in grade A hospitals. The awareness rate, treatment rate and control rate of hypertension and hyperlipidemia were even lower in primary hospitals (35). The difference in clinical outcomes may be the underlying reason why patients choose a grade A hospital instead of a primary medical institution. As some studies have shown, medical treatment in China is the mode of individual free choice, and whether patients can be cured is the primary factor that patients consider when making medical choices (13).

The findings in this study will facilitate us to formulate policies and effective measures for the hierarchical diagnosis and treatment of hypertensive patients (36). Improving the effectiveness of hypertension diagnosis and treatment in primary medical institutions will be the goal, which can be achieved by increasing the training of medical staff, formulating standardized diagnosis and treatment procedures, digitizing the management of patients' blood pressure data, and AI checking the accuracy of prescriptions.

This study has some limitations, such as: the study data are obtained from a structured questionnaire. In the future, semi-structured interviews will provide better insight into the factors and perceptions of patients' health seeking behaviors. The reliability of the questionnaire can be further evaluated using the Cronbach's alpha. And a standard quality of life assessment questionnaire will provide more information. To expand the variety of diseases in the attending patients to perform a multi-level aggregation analysis will provide more refined evidence for policy making.

Conclusion

The choices of medical behavior of patients with cardiovascular disease risk factors are not only influenced by socioeconomic factors but also the differences in clinical outcomes of treatment which might be the underlying cause especially for chronic diseases such as hypertension.

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/s.

Ethics statement

The studies involving humans were approved by the Ethics Committee of Beijing Anzhen Hospital affiliated to Capital Medical

University (IRB No. 2013024). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

LG: Writing – original draft, Project administration. XD: Conceptualization, Writing – review & editing. HW: Data curation, Writing – review & editing. SX: Formal analysis, Writing – review & editing. JD: Data curation, Writing – review & editing. XK: Data curation, Writing – review & editing. XY: Project administration, Writing – review & editing. CW: Visualization, Writing – review & editing. JZD: Project administration, Writing – review & editing. CM: Resources, Writing – review & editing. LE: Writing – original draft, Writing – review & editing.

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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.

Publisher's note

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Association of geographical disparities and segregation in regional treatment facilities for Black patients with aneurysmal subarachnoid hemorrhage in the United States

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Background and objectives: This study investigates geographic disparities in aneurysmal subarachnoid hemorrhage (aSAH) care for Black patients and aims to explore the association with segregation in treatment facilities. Understanding these dynamics can guide efforts to improve healthcare outcomes for marginalized populations.

Methods: This cohort study evaluated regional differences in segregation for Black patients with aSAH and the association with geographic variations in disparities from 2016 to 2020. The National Inpatient Sample (NIS) database was queried for admission data on aSAH. Black patients were compared to White patients. Segregation in treatment facilities was calculated using the dissimilarity (D) index. Using multivariable logistic regression models, the regional disparities in aSAH treatment, functional outcomes, mortality, and end-of-life care between Black and White patients and the association of geographical segregation in treatment facilities was assessed.

Results: 142,285 Black and White patients were diagnosed with aSAH from 2016 to 2020. The Pacific division (D index = 0.55) had the greatest degree of segregation in treatment facilities, while the South Atlantic (D index = 0.39) had the lowest. Compared to lower segregation, regions with higher levels of segregation (global F test $p < 0.001$) were associated a lower likelihood of mortality (OR 0.91, 95% CI 0.82–1.00, $p = 0.044$ vs. OR 0.75, 95% CI 0.68–0.83, $p < 0.$ 001) ($p =$ 0.049), greater likelihood of tracheostomy tube placement (OR 1.45, 95% CI 1.22–1.73, $p < 0.001$ vs. OR 1.87, 95% CI 1.59–2.21, $p < 0.001$) ($p < 0.$ 001), and lower likelihood of receiving palliative care (OR 0.88, 95% CI 0.76–0.93, $p < 0.001$ vs. OR 0.67, 95% CI 0.59–0.77, $p < 0.001$) ($p = 0.029$).

Conclusion: This study demonstrates regional differences in disparities for Black patients with aSAH, particularly in end-of-life care, with varying levels of segregation in regional treatment facilities playing an associated role. The

findings underscore the need for targeted interventions and policy changes to address systemic healthcare inequities, reduce segregation, and ensure equitable access to high-quality care for all patients.

KEYWORDS

regional disparities, Black patients, aneurysmal subarachnoid hemorrhage, segregation, healthcare disparities, equitable care

Introduction

Black patients in America face significant challenges within the healthcare system, resulting in disparities that manifest as higher rates of chronic diseases, limited access to quality healthcare, and worse health outcomes compared to their White counterpart (1, 2). These disparities are evidenced by elevated rates of chronic diseases, restricted access to high-quality healthcare, and poorer health outcomes among Black patients compared to their White counterparts (2–8). Moreover, these disparities extend beyond general health conditions to acute medical crises, notably in neurosurgical emergencies like aneurysmal subarachnoid hemorrhage (aSAH). For instance, studies have indicated that Black patients with aSAH experience higher mortality rates and greater disability compared to White patients (9–11).

The root causes of these health disparities are multifaceted. They include limited access to healthcare, socioeconomic inequalities, cultural barriers, implicit biases, and disparities in the quality of care (3, 12–14). These factors are further compounded by geographical variations, with differences in healthcare infrastructure, socioeconomic conditions, cultural norms, healthcare provider demographics, and historical patterns of segregation (5, 6, 15). Segregation in healthcare refers to the unequal distribution of medical services, resources, and opportunities based on factors such as race, ethnicity, socioeconomic status, or geographic location. It can lead to significant disparities in access to quality medical services and resources, resulting in marginalized communities receiving substandard care and experiencing worse health outcomes. Hobar et al. explicated segregation in regional neonatal intensive care units (NICU) among Black and White newborns, with Black babies being treated at lower quality NICUs than White neonates (3, 16).

The legacy of segregation in healthcare for Black Americans is marked by a history of systemic discrimination and exclusion. In the Jim Crow era, legally sanctioned racial segregation relegated Black patients to separate, "colored" facilities, which were typically plagued by chronic underfunding and a dearth of resources, resulting in care that was markedly inferior to that available in institutions serving White patients. These "Negro hospitals," as they were then known, became symbols of the broader injustices of the time—enduring emblems of inequality in American healthcare (17–22). Although the overt legal structures of segregation have been dismantled, the shadows of these historical disparities continue to loom over contemporary healthcare outcomes for Black patients (19–22).

It is within this context that our study examines the relationship between the continued segregation in healthcare settings and its impact on the treatment and outcomes of Black patients suffering from aSAH. Existing research indicates that minority groups with aSAH, including Black patients, often receive more aggressive medical interventions, like tracheostomy and gastrostomy tube placements, as well as blood transfusions, than their White counterparts (23). In contrast, these patients are less frequently involved in palliative care consultations or designated with Do Not Resuscitate (DNR) status (23). We hypothesize a direct correlation between the degree of segregation in treatment facilities and the observed disparities in care. Specifically, we focus on the prevalence of aggressive treatment interventions and the lack of engagement with palliative care services and DNR status in areas with higher segregation indices. By shedding light on the pervasive nature of these disparities and their association with segregation, this study seeks to articulate the ongoing challenges in achieving healthcare equity. Furthermore, it aspires to lay the groundwork for interventions specifically designed to counteract these disparities and foster a more equitable healthcare landscape for Black patients with aSAH.

Methods

Data source

The National Inpatient Sample (NIS) database was queried from 2016 to 2020 for diagnosis of ruptured aSAH using the International Classification for Disease version 10 (ICD-10). Patients with ICD - 10 codes 160.00–160.09 met the primary inclusion criteria (Supplementary Table S1). Patients with traumatic SAH ($n = 1,740$) or SAH associated with an arteriovenous malformation ($n = 23,575$) were excluded from the study (Supplementary Figure S1). This approach aligns with the study's aim to investigate healthcare disparities and outcomes in aSAH, distinct in its pathophysiology and treatment from conditions like traumatic SAH and SAH due to arteriovenous malformations. By concentrating on aSAH, our study aligns with existing literature and avoids the confounding variables introduced by the inclusion of other SAH etiologies, thereby ensuring a more accurate and homogenous examination of the specific disparities and outcomes in this patient group. The NIS, part of the Healthcare Cost and Utilization Project (HCUP), is a vast database containing de-identified inpatient hospitalization data from a wide array of U.S. hospitals, enhancing its national representativeness. It utilizes discharge weights from participating hospitals to provide nationally representative estimates, facilitating diverse healthcare research. However, limitations include potential billing and coding inaccuracies, variations in hospital reporting practices, and the exclusion of certain facilities like federal hospitals. Additionally, its focus is solely on inpatient data, excluding outpatient care. Due to the de-identified, retrospective nature of this study, Institutional Review Board approval was not sought.

Population

The NIS database stratifies race into 6 groups: White, Black, Hispanic, Asian/Pacific Islander, Native American, and Other. Only patients listed as Black or White were included in this study. Our analysis focuses on Black and White patients to closely examine the significant disparities predominantly observed between these racial groups. This targeted approach not only captures a vital element of the wider discourse on racial inequalities in healthcare but also acknowledges the enduring impact of historical segregation and systemic biases. These long-standing issues have disproportionately affected Black communities in the United States, perpetuating a cycle of healthcare disparities. Patients were further divided by geographical location using NIS-provided United States Census divisions (New England, Middle Atlantic, East North Central, West North Central, South Atlantic, East South Central, West South Central, Mountain, and Pacific). Supplementary Table S2 outlines the corresponding states that make up each U.S. Census division.

Outcomes and covariates

Research questions centered on treatment disparities, functional outcomes, mortality, and end-of-life care. Treatment was dichotomized into two groups for patients undergoing aneurysm treatment (via open surgical clipping or endovascular therapy) and patients who did not receive either treatment. Functional outcomes in SAH cases were assessed using the NIS-SAH Outcomes Measure (NIS-SOM), a validated instrument provided by the NIS data source. This comprehensive metric evaluates treatment effectiveness and forecasts overall patient prognosis by integrating clinical, demographic, and hospital-related variables, similar to the approach used in modified Rankin scores. The NIS-SOM, is a dichotomous tool that classifies patient outcomes post-discharge into two categories: "good outcome" and "poor outcome." A "good outcome" signifies a patient's discharge to their home or a rehabilitation facility, reflecting a positive recovery path. In contrast, a "poor outcome" includes a spectrum of less favorable scenarios, such as in-hospital mortality, or discharge to a facility offering nursing care, extended care, long-term acute care, or hospice services. The design of the NIS-SOM aims to efficiently delineate patient recovery levels and care needs at the point of discharge, offering a clear and structured measure for evaluating SAH patient outcomes (24). To investigate disparities in end-of-life care, we examined the racial differences in life-sustaining interventions (mechanical ventilation, tracheostomy tube placement, gastrostomy tube placement, and blood transfusions), the utilization of palliative care services, and DNR. All analyses included the following covariates: age, sex (male and female), admission year, hospital size, teaching status, primary expected payer (Medicare, Medicaid, private insurance, self-pay, no charge, other), hypertension, obesity, smoking status, coronary artery disease, chronic kidney disease, hyperlipidemia, diabetes mellitus, alcohol abuse, atrial fibrillation, and the NIS SAH Severity Score (NIS-SSS) (25, 26). The NIS-SSS is a scoring system derived from the NIS database to assess the severity of aSAH using clinical and demographic data. Validated against established grading systems like the Hunt and Hess scale, it provides a reliable tool for stratifying patients and analyzing outcomes in aSAH studies (27). Outcomes and covariates not directly provided by the NIS were derived using secondary discharge diagnoses in the data source (Supplementary Table S2).

Measure of segregation

To measure segregation in treatment facilities between Black and White patients within each U.S. Census region, we calculated the Duncan dissimilarity (D) index as outlined by Austin et al. (7). The *D* index is a statistical measure used to quantify the degree of segregation between two groups within a geographic area. It measures the proportion of individuals from one group who would need to change their location to achieve an even distribution with the other group. The *D* index ranges from zero to one, where zero indicates complete integration or no segregation, and one represents complete segregation. The *D* index was chosen for this study due to its established reliability, as demonstrated by its application by the U.S. Census Bureau for assessing residential segregation, and its successful use in previous research to quantify racial segregation in healthcare facilities, making it an ideal measure for exploring segregation's impact on healthcare disparities (7, 28–30). The formula used for calculating the *D* index along with an example calculation is illustrated in Figure 1. *D* indexes were calculated by U.S. census region for each of the studied years.

Statistical analysis

Univariate analysis was performed to explore differences in rates of mechanical ventilation, tracheostomy tube placement, gastrostomy tube placement, blood transfusions, the utilization of palliative care services, and DNR status for race (Black vs. White) and region (Supplementary Table S4). For variables with $p < 0.20$, multivariable logistic regression models were performed to control for covariate influence. All multivariable models used the above-outlined covariates. Patients missing data on variables of interest were excluded from the analyses. Racial disparities were compared to the national average for each of the nine census divisions. Analysis of variance (ANOVA) was used to compare differences in the level of segregation as measured by the *D* index among the regions. Finally, the impact of the level of segregation on the outcomes of interest was tested by comparing high (*D* index of 0.50 or higher) and low *D* index (*D* index <0.50) regions using multivariable logistic regression with the included covariates followed by a global *F* test when both regions showed statistical significance. If the overall *F* test was statistically significant, then high and low *D* index regions were compared using a *t*-test for the outcome of interest. All tests for significance were two-sided, with a *p*-value of 0.05 or less defined as statistically significant. This threshold of 0.05 was chosen to align with the convention used in other publications utilizing the NIS Database, ensuring consistency in our approach and facilitating comparisons with similar studies (23). Statistical analysis was performed using R version 4.2.2. This study adheres to The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) initiative recommendations (31).

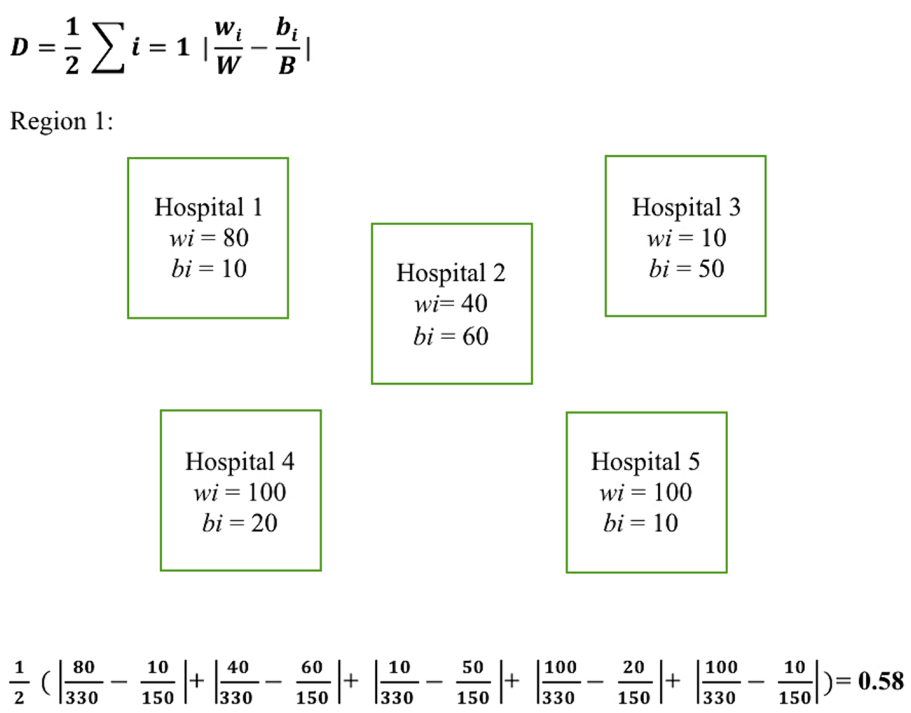

FIGURE 1
Dissimilarity index equation with example calculation. Illustrates the equation used for calculating the *D* index along with a sample calculation. Here, w_i and b_i represent the number of White and Black patients treated at a given hospital within a region, respectively. *W* and *B* denote the total number of White and Black patients respectively within the region that the *D* index is being calculated.

Results

We included 142,285 (113,060 White, 29,255 Black) patients hospitalized with aSAH from 2016 to 2020 in this study. Most Black patients in the cohort were cared for in the South Atlantic (33%), East North Central (16%), and Middle Atlantic (15.90%) regions. Similar observations were noted with White patients: South Atlantic (20%), East North Central (18%), and Middle Atlantic (13%). Table 1 provides a summary of baseline characteristics for the study cohort.

Regional disparities in treatment, functional outcomes, and mortality

Nationally, Black patients had higher treatment rates following aSAH in comparison to White patients (OR 1.10, 95% CI 1.02–1.18, $p = 0.013$). However, significant racial differences in undergoing aSAH treatment were only demonstrated in the East South-Central region (OR 1.52, 95% CI 1.24–1.88, $p < 0.001$). Black patients had worse NIS-SOM outcomes on the national level than White patients (OR 1.10, 95% 1.02–1.19, $p = 0.010$), with significantly worse functional outcomes seen in the Middle Atlantic (OR 1.27, 95% CI 1.04–1.55, $p = 0.022$) and West South-Central regions (OR 1.27, 95% CI 1.03–1.56, $p = 0.025$). Nationally, the mortality rate for Black patients was lower than that of White patients following aSAH (OR 0.81, 95% CI 0.75–0.88, $p < 0.001$). Black patients in the East North Central (OR 0.68, 95% CI 0.55–0.83, $p < 0.001$), East South Central (OR 0.63, 95% CI 0.47–0.84, $p = 0.002$), and West South Central (OR 0.74, 95% CI 0.58–0.96, $p = 0.022$) demonstrated statistically significant lower likelihood of mortality.

Geographical variance in disparities in end-of-life care

Black patients were twice as likely as White patients to have tracheostomy tube placement in the Pacific division (OR 2.17, 95% CI 1.46–3.22, $p < 0.001$). Black patients in the Middle Atlantic (OR 1.76, 95% CI 1.27–2.43, $p < 0.001$), East North Central (OR 1.83, 95% CI 1.39–2.42, $p < 0.001$), and West South Central (OR 1.83, 95% CI 1.39–2.42, $p < 0.001$) regions had higher likelihood of tracheostomy tube placement in comparison to the national level (OR 1.67, 95% CI 1.48–1.89 $p < 0.001$). Conversely, a lower likelihood of tracheostomy placement than the national average was seen in the South Atlantic (OR 1.33, 95% CI 1.05–1.68, $p = 0.018$) and East South Central (OR 1.61, 95% CI 1.10–2.35, $p = 0.015$) regions. Black patients in the New England region had the greatest likelihood of gastrostomy tube placement (OR 2.00, 95% CI 1.13–3.33, $p = 0.017$), while having the least likelihood of receiving palliative care services (OR 0.42, 95% CI 0.22–0.80, $p = 0.008$) and utilization of DNR (OR 0.47, 95% CI 0.27–0.85, $p = 0.012$). Black patients in the South Atlantic had the lowest degree of disparity in the likelihood of gastrostomy tube placement (OR 1.36, 95% CI 1.14–1.61, $p = 0.001$) and receipt of palliative care (OR 0.76, 95% CI 0.65–0.89, $p = 0.001$). Table 2 summarizes the disparities for Black patients compared to White patients by region in treatment, outcomes, mortality, and end-of-life care.

TABLE 1 Patient and hospital demographics by race.

Characteristic		White		Black	
		No.	(%)	No.	(%)
Total number of patients		113,060	79	29,225	21
Sex					
	Male	50,410	45	11,915	41
	Female	62,650	55	17,310	59
Region					
	New England	6,315	6	690	2
	Middle Atlantic	14,570	13	3,970	14
	East North Central	19,835	18	4,665	16
	West North Central	7,860	7	980	3
	South Atlantic	22,400	20	9,765	33
	East South Central	9,375	8	2,835	10
	West South Central	10,365	9	3,420	12
	Mountain	8,025	7	600	2
	Pacific	14,315	13	2,300	8
Comorbidity					
	Chronic kidney disease	12,595	11	5,410	19
	Atrial fibrillation	185	0.16	15	0.05
	Coronary artery disease	18,290	16	2,985	10
	Alcohol abuse	2,550	2	790	3
	Hypertension	60,670	54	14,860	51
	Diabetes mellitus	22,495	20	7,645	26
	Obesity	12,885	11	4,510	15
	Hyperlipidemia	39,280	35	7,975	27
	Congestive heart failure	14,375	13	4,535	16
	Smoking	23,145	20	4,555	16
	Admission GCS < 8	4,635	4	1,280	4
Bed size					
	Small	10,550	9	2,565	9
	Medium	24,925	22	6,795	23
	Large	77,585	69	19,865	68
Primary expected payer					
	Medicare	60,395	53	11,555	40
	Medicaid	11,530	10	6,670	23
	Private insurance	33,380	30	7,860	27
	Self-pay	4,245	4	2,080	7
	No charge	230	0.20	175	0.60
	Other	3,280	3	885	3

Association of racial segregation in regional treatment facility with disparities

Segregation between White and Black patients in treatment facilities following aSAH was greatest in the Pacific (*D* index = 0.55) and Mountain (*D* index = 0.54) divisions, and lowest in the South Atlantic (*D* index = 0.39) and West South Central (*D* = 0.46) divisions. Slight variance in the *D* index within a region were noted over the five-year study, as illustrated in Supplementary Figure S2. Inter-regional comparison showed that the difference in *D* index was statistically significant across all regions over the studied period excluding comparisons between the New England and Middle Atlantic regions (Supplementary Table S3). Since 2012, the NIS has ceased providing hospital names or identifiers, offering only deidentified hospital IDs and limited characteristics such as teaching status, bed size, and hospital type. Consequently, crucial metrics for assessing hospital quality are missing. This limitation impacts our study's use of the *D* index, as it restricts our ability to differentiate between patients treated at lower-quality and higher-quality facilities when analyzing segregation in treatment facilities.

Black patients in regions with higher levels of segregation had a higher likelihood of poor functional outcomes compared to White patients (OR 1.15, 95% CI 1.04–1.29, $p = 0.008$). Conversely, no statistically significant difference was noted in likelihood of poor functional outcomes between Black and White patients in low *D* index regions (OR 1.07, 95% CI 0.97–1.19, $p = 0.181$). The likelihood of mortality by *D* index level (global *F* test $p < 0.001$) was higher in less segregated (OR 0.91, 95% CI 0.82–1.00, $p = 0.044$) compared to more segregated (OR 0.75, 95% CI 0.68–0.83, $p < 0.001$) regions ($p = 0.049$). Notable differences were also seen when comparing low and high *D* index regions (global *F* test $p < 0.001$) for tracheostomy tube placement (OR 1.45, 95% CI 1.22–1.73, $p < 0.001$ vs. OR 1.87, 95% CI 1.59–2.21, $p < 0.001$) ($p < 0.001$) and receipt of palliative care (OR 0.88, 95% CI 0.76–0.93, $p < 0.001$ vs. OR 0.67, 95% CI 0.59–0.77, $p < 0.001$) ($p = 0.029$). No statistically significant difference was found when comparing disparities between low and high *D* index region (global *F* test $p < 0.001$) for gastrostomy tube placement (OR 1.62, 95% CI 1.12–2.36, $p = 0.011$ vs. OR 1.68, 95% CI 1.46–1.93, $p < 0.001$) ($p = 0.783$), blood transfusions (OR 1.44, 95% CI 1.20–1.74, $p < 0.001$ vs. OR 1.49, 95% CI 1.24–1.79, $p < 0.001$) ($p = 0.738$), and DNR status (OR 0.68, 95% CI 0.61–0.77, $p < 0.001$ vs. OR 0.67, 95% CI 0.59–0.75, $p < 0.001$) ($p = 0.841$). The relationship between the *D* index and likelihood of tracheostomy tube placement and receiving palliative care is represented as a heatmap in Figure 2. Table 3 provides a summary of all odds ratio calculations by high versus low *D* index region.

Discussion

We observed Black patients nationally underwent treatment at a slightly higher rate—and despite worse functional outcomes, Black patients were less likely to have inpatient mortality than White patients following aSAH. While the higher rate of treatment and lower mortality would at first seem to represent a positive finding, it must be taken in context. Black patients were more likely on the national level to have tracheostomy tube placement, gastrostomy tube placement, and receive blood transfusion. Black patients were less likely to have palliative care involvement or code status changed to DNR. These findings reflect a disparity in end-of-life care in that Black patients have a lower mortality and higher treatment rate because they are undergoing more life-sustaining care and less palliation in situations where the outcomes are worse. Cruz-Flores et al. report similar findings in a NIS study on intra-cerebral hemorrhages (ICH) that demonstrated Black patients with ICH are more likely to utilize lifesaving (surgical intervention), life prolonging (mechanical

TABLE 2 Odds ratios for treatment, functional outcomes, and end-of-life care for Black patients by region.

	Treatment			NIS-SOM		
Region	OR	95% CI	*p*-value	OR	95% CI	*p*-value
New England	0.74	0.43–1.27	0.272	1.50	0.96–2.34	0.075
Middle Atlantic	1.16	0.94–1.43	0.177	1.27	1.04–1.55	**0.022**
East North Central	0.89	0.75–1.06	0.180	1.16	0.94–1.43	0.178
West North Central	1.04	0.70–1.54	0.842	0.89	0.54–1.48	0.663
South Atlantic	1.05	0.91–1.20	0.504	1.01	0.88–1.15	0.903
East South Central	1.52	1.24–1.88	**<0.001**	1.14	0.88–1.47	0.316
West South Central	1.17	0.94–1.45	0.156	1.27	1.03–1.56	**0.025**
Mountain	0.67	0.40–1.12	0.124	0.89	0.60–1.31	0.543
Pacific	1.15	0.91–1.44	0.235	1.04	0.83–1.31	0.725
National	1.10	1.02–1.18	**0.013**	1.10	1.02–1.19	**0.010**

	Mortality			Mechanical ventilation		
Region	OR	95% CI	*p*-value	OR	95% CI	*p*-value
New England	1.28	0.81–2.03	0.288	9825.19	28.35–3405687.98	**0.002**
Middle Atlantic	0.89	0.72–1.10	0.277	1.20	0.49–2.94	0.686
East North Central	0.68	0.55–0.83	**< 0.001**	1.85	1.07–3.21	**0.028**
West North Central	1.06	0.66–1.68	0.813	0.06	0.01–0.50	**0.010**
South Atlantic	0.98	0.85–1.13	0.781	1.52	0.64–3.59	0.34
East South Central	0.63	0.47–0.84	**0.002**	1.7	0.83–3.49	0.149
West South Central	0.74	0.58–0.96	**0.022**	0.96	0.30–3.05	0.943
Mountain	0.67	0.36–1.22	0.188	1.7	0.51–5.67	0.39
Pacific	0.79	0.61–1.01	0.059	0.92	0.33–2.56	0.868
National	0.81	0.75–0.88	**<0.001**	1.3	0.89–1.89	0.172

	Tracheostomy			Gastrostomy		
Region	OR	95% CI	*p*-value	OR	95% CI	*p*-value
New England	1.84	0.86–3.93	0.115	2.00	1.13–3.53	**0.017**
Middle Atlantic	1.76	1.27–2.43	**0.001**	1.89	1.43–2.51	**< 0.001**
East North Central	1.83	1.39–2.42	**< 0.001**	1.66	1.31–2.11	**<0.001**
West North Central	1.38	0.72–2.66	0.328	1.50	0.89–2.54	0.128
South Atlantic	1.33	1.05–1.68	**0.018**	1.36	1.14–1.61	**0.001**
East South Central	1.61	1.10. - 2.35	**0.015**	1.70	0.83–3.49	0.149
West South Central	1.86	1.29–2.66	**0.001**	1.95	1.44–2.63	**< 0.001**
Mountain	1.95	0.93–4.06	0.075	0.98	0.49–1.96	0.945
Pacific	2.17	1.46–3.22	**< 0.001**	1.52	1.11–2.07	**0.009**
National	1.67	1.48–1.89	**<0.001**	1.58	1.43–1.74	**<0.001**

	Transfusions			Palliative care		
Region	OR	95% CI	*p*-value	OR	95% CI	*p*-value
New England	0.69	0.27–1.74	0.428	0.42	0.22–0.80	**0.008**
Middle Atlantic	1.61	1.19–2.20	**<0.001**	0.78	0.61–1.01	0.059
East North Central	1.37	0.96–1.93	0.079	0.67	0.53–0.84	**0.001**
West North Central	2.38	0.95–5.95	0.064	0.76	0.46–1.23	0.261
South Atlantic	1.23	0.98–1.55	0.072	0.76	0.65–0.89	**0.001**

(Continued)

TABLE 2 (Continued)

	Transfusions			Palliative care		
Region	OR	95% CI	*p*-value	OR	95% CI	*p*-value
East South Central	1.74	1.06–2.86	**0.029**	0.59	0.43–0.80	**0.001**
West South Central	1.79	1.18–2.70	**0.006**	0.67	0.51–0.87	**0.003**
Mountain	1.81	0.83–3.95	0.135	0.57	0.32–1.02	0.059
Pacific	1.25	0.82–1.90	0.304	0.68	0.51–0.91	**0.008**
National	1.49	1.30–1.70	**<0.001**	0.69	0.63–0.75	**<0.001**

	Do not resuscitate		
Region	OR	95% CI	*p-value*
New England	0.47	0.27–0.85	**0.012**
Middle Atlantic	0.75	0.59–0.96	**0.02**
East North Central	0.60	0.49–0.96	**0.001**
West North Central	0.71	0.46–1.09	0.116
South Atlantic	0.70	0.60–0.82	**<0.001**
East South Central	0.55	0.42–0.73	**<0.001**
West South Central	0.71	0.55–0.91	**0.008**
Mountain	0.52	0.30–0.91	**0.019**
Pacific	0.75	0.57–0.99	**0.044**
National	0.66	0.61–0.72	**<0.001**

NIS-SOM, National Inpatient Sample subarachnoid hemorrhage outcomes measure. Treatment refers to patients receiving aneurysm securement via surgical clipping or endovascular coiling. The bold values represent statistically significant values.

ventilation, tracheostomy tube, gastrostomy tube, and blood transfusions) interventions, and less likely to receive palliative and hospice care compared to White patients. These findings are posited to reflect a lower likelihood of mortality for Black patients with ICH (32).

Several explanatory factors have been cited for this global disparity within healthcare in aggressive end-of-life care (33–35). One factor is implicit bias, which refers to unconscious attitudes and stereotypes that can influence medical decision-making. Research has shown that healthcare professionals, including doctors, may hold implicit biases that contribute to perceiving Black patients as having less sensitive nerve endings and thicker skin, or feeling less empathy toward their pain, leading to the belief that they can tolerate more aggressive treatments (35, 36). Additionally, historical mistrust stemming from past instances of medical mistreatment and experimentation on Black communities can create a fear of being under-treated, prompting both patients and healthcare providers to opt for more aggressive interventions (37–39). Furthermore, disparities in access to quality healthcare and socioeconomic factors, such as limited health insurance coverage and fewer healthcare resources in predominantly Black communities, can contribute to delayed or inadequate care requiring more aggressive interventions for more severe conditions at the time of clinical presentation (40–43).

Cultural and religious considerations are pivotal in healthcare decisions, especially at the end of life. A study examining advanced cancer patients revealed that those whose spiritual needs were met by their healthcare team were more inclined to utilize hospice services and less likely to seek aggressive treatments, underscoring the significance of spirituality in these decisions (44). Interestingly, this finding was not race-specific. In contrast, a study focusing specifically on race found that while religiosity influenced the use of DNR orders among White cancer patients, it did not hold the same sway for Black patients (45). Additionally, no study to date demonstrates that Black patients rely more heavily on religious guidance than White patients when making end-of-life choices. This points to a crucial insight: while spiritual beliefs are indeed a factor, they do not singularly drive the decision-making process within the Black community, which is marked by a rich diversity of beliefs and practices shaped by personal, familial, and regional distinctions. Overstating the role of culture and religion may risk simplifying the complex interplay of influences on healthcare outcomes and divert attention from systemic barriers such as structural racism and healthcare inequities. Therefore, it's essential to view cultural and religious beliefs as part of a wider array of determinants that collectively influence healthcare outcomes, rather than as isolated or predominant factors.

While our study found that disparities are present throughout every geographic location in the country, some regions had much higher disparities. Black patients in the Northeast (New England and Middle Atlantic) and West (Mountain and Pacific) tend to have greater disparities in life-sustaining interventions, use of palliative care, and DNR status. The South Atlantic division had the least disparities. Various factors may contribute to the observed regional variances. In this study, we demonstrated the association between disparities and segregation in treatment facilities by elucidating a relationship with disparities in treatment and clinical outcome. Segregation between Black and White patients with aSAH was greatest in regions with the largest disparities, i.e., Northeast and West. When comparing high and low segregation, more segregated regions were associated with worse

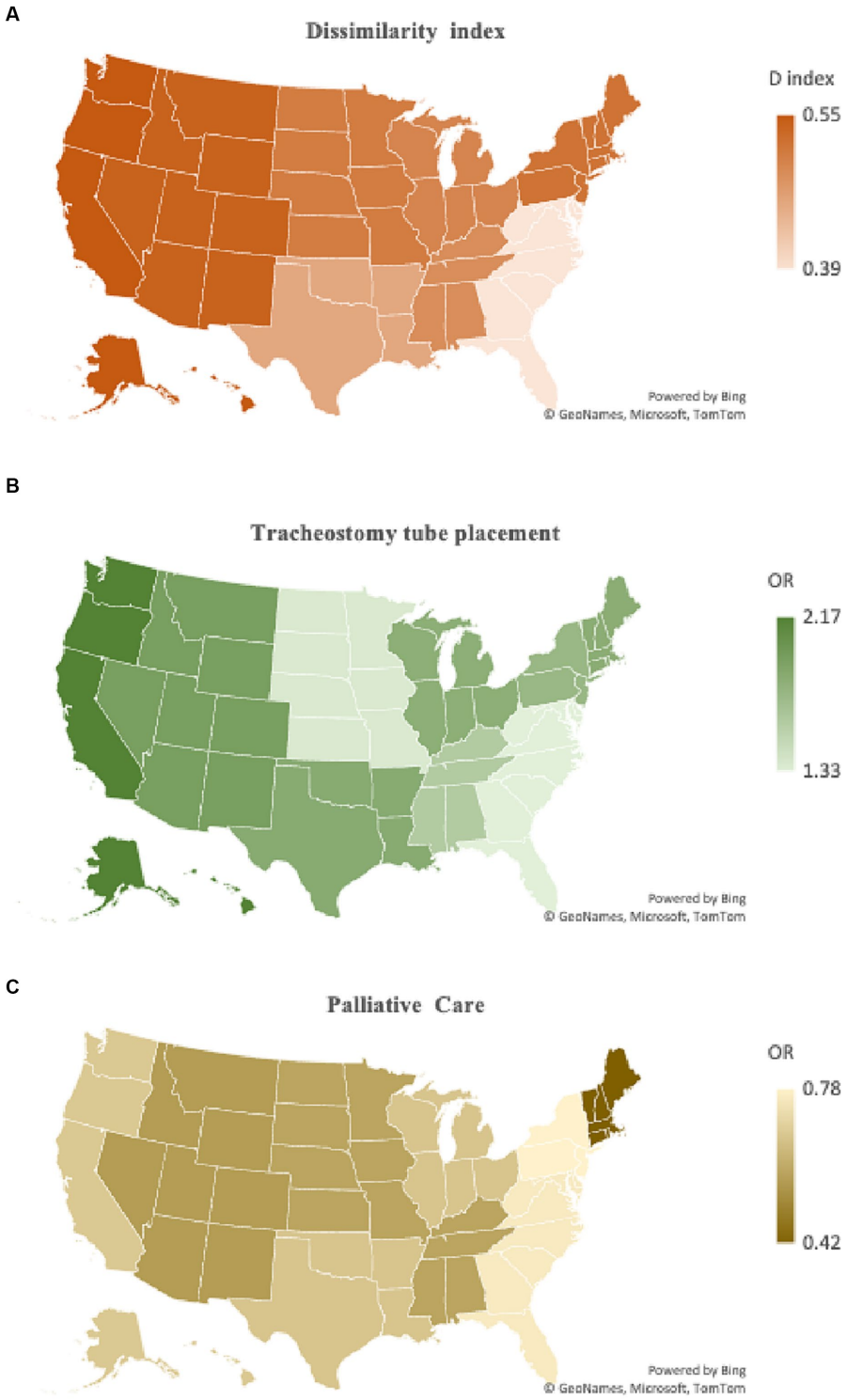

FIGURE 2
Geographical variation in dissimilarity index and association with likelihood of tracheostomy tube placement and receipt of palliative care. **(A)** A heatmap of dissimilarity index by U.S. Census region is juxtaposed to **(B)** a heatmap representation of likelihood of tracheostomy tube placement and **(C)** palliative care use for Black patients following aSAH. Regions with higher levels of segregation also had higher levels of tracheostomy tube placement for Black patients and lower palliative care consultation. This visually demonstrates the association of levels of segregation and disparities some aspects of end-of-life care.

functional outcomes and a lower likelihood of mortality, but notably higher likelihood of tracheostomy tube placement and decreased utilization of palliative care services.

The persistence of segregation for Black patients in treatment facilities is an amalgam of a complex interplay of historical, social, and systemic factors. Deep-rooted racial biases, discriminatory practices,

TABLE 3 Odds Ratios for treatment, functional outcomes, morality, and end-of-life care by high versus low *D* index Region.

	Treatment			NIS-SOM			Mortality		
	OR	95% CI	*p*-value	OR	95% CI	*p*-value	OR	95% CI	*p*-value
High *D* index Region	1.02	0.92–1.14	0.689	1.15	1.04–1.29	**0.008**	0.75	0.68–0.83	**<0.001**
Low *D* index Region	1.14	1.03–1.26	**0.010**	1.07	0.97–1.19	0.181	0.91	0.82–1.00	**0.044**

	Mechanical ventilation			Tracheostomy			Gastrostomy		
	OR	95% CI	*p*-value	OR	95% CI	*p*-value	OR	95% CI	*p*-value
High *D* index Region	1.10	0.65–1.85	0.725	1.87	1.59–2.21	**<0.001**	1.68	1.46–1.93	**<0.001**
Low *D* index Region	1.44	0.85–2.46	0.180	1.45	1.22–1.73	**<0.001**	1.62	1.12–2.36	**0.011**

	Palliative care			Do not resuscitate			Transfusions		
	OR	95% CI	*p*-value	OR	95% CI	*p*-value	OR	95% CI	*p*-value
High *D* index Region	0.67	0.59–0.77	**<0.001**	0.66	0.59–0.75	**<0.001**	1.49	1.24–1.79	**<0.001**
Low *D* index Region	0.88	0.76–0.93	**<0.001**	0.68	0.61–0.77	**<0.001**	1.44	1.20–1.74	**<0.001**

The bold values represent statistically significant values.

and structural inequalities have perpetuated the unequal distribution of resources and opportunities within the healthcare system (46–48). Historical patterns of racial segregation, such as redlining and discriminatory housing practices, have led to the concentration of Black populations in marginalized neighborhoods with limited access to quality healthcare facilities (46–48). This spatial segregation, combined with socioeconomic disparities and inadequate healthcare infrastructure, has created barriers to equal treatment and access to care for Black individuals (49–52). Additionally, implicit biases and stereotypes among healthcare providers may contribute to differential treatment and perpetuate disparities in the delivery of healthcare services (53). Furthermore, Black patients seeking care are less likely to be transferred to a different treatment facility compared to White patients, including patients being treated for aSAH (54, 55).

To bridge the healthcare gap and combat systemic barriers, a comprehensive strategy is vital for ensuring equitable access to treatment for Black individuals, thus reducing disparities. This begins with healthcare systems conducting in-depth evaluations of facility placement and service availability, especially in highly segregated areas. Such assessments should aim to uncover and rectify care deficiencies, potentially through the strategic establishment of new facilities or the enhancement of services in existing ones within underserved Black communities.

Furthermore, forging partnerships with community organizations can improve healthcare system navigation for Black patients. Continuous bias training for healthcare providers is imperative, fostering an environment of ongoing education to combat unconscious biases that may influence patient care. In addition to this, it is crucial for healthcare professionals to embrace shared decision-making, honoring the cultural and individual preferences of Black patients, with a keen focus on end-of-life care choices. Policy reform should also focus on decentralizing high-quality care, ensuring a fair distribution of medical resources and enhancing patient transfer protocols, so that every patient, regardless of race, has access to the best possible care. Finally, culturally sensitive public health initiatives are necessary. These should provide education and resources that resonate with the Black community's varied values and beliefs, particularly around end-of-life care. Public health campaigns can also play a role in raising awareness about the benefits of advance care planning within these communities. By implementing these steps, we can begin dismantling the deep-rooted barriers contributing to healthcare disparities, paving the way for a future where equitable care is not an ideal, but a reality for all patients.

Limitations

Our study, while contributing important findings, is subject to several notable limitations that should be considered. A primary limitation stems from the nature of the NIS data source. Since 2012, the NIS no longer provides specific hospital names or identifiers, limiting our ability to assess the impact of individual hospital characteristics on treatment outcomes. This absence of detailed identifiers could potentially affect the external validity of our findings, as it restricts our capacity to generalize results to specific types of hospitals or geographical locations.

Furthermore, while the NIS offers a vast array of data, it primarily consists of administrative records. This reliance on administrative data can lead to a lack of nuanced clinical details and may introduce inaccuracies due to coding errors. Additionally, the large sample sizes typical of NIS data, though beneficial for statistical power, do not inherently imply clinical significance. In cases where statistical differences do not reflect clinically meaningful distinctions, the practical applicability of our findings may be limited. Hence,

interpretations of our results should be made with an understanding that both statistical and clinical significance are crucial for drawing comprehensive and applicable conclusions.

Conclusion

Nationally and regionally, our study found that Black patients undergoing aSAH treatment are more frequently subjected to life-sustaining interventions such as tracheostomy, gastrostomy, and blood transfusions, yet they receive less palliative care and fewer code status changes to DNR. This occurs despite them experiencing poorer functional outcomes, highlighting a significant disparity in treatment approaches and end-of-life care decisions Notably, the Northeast and West regions exhibited the most significant treatment disparities, correlating with the highest levels of racial segregation in healthcare facilities, while the South Atlantic division showed the least. This pattern suggests that areas with higher segregation see more pronounced disparities in both treatment and clinical outcomes.

Previous presentation/publication

The abstract was presented at the Congress of Neurological Surgeons (CNS) annual meeting in September 2023 and awarded the CNS foundation diversity, equity, and inclusion abstract award for 2023. No parts of this study have otherwise been presented or published elsewhere.

Data availability statement

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

Author contributions

J-LK: Writing – review & editing, Writing – original draft, Visualization, Validation, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. LF: Visualization, Software, Resources, Investigation, Formal analysis, Writing – review & editing, Validation, Methodology, Data curation. AB: Writing – original draft, Visualization. FD: Data curation, Writing – review & editing, Visualization, Validation. MB: Writing – review & editing, Validation, Writing – original draft, Visualization. AH: Writing – review & editing, Visualization, Validation. AR: Software, Methodology, Formal analysis, Conceptualization, Writing – review & editing, Visualization, Validation, Data curation. JP: Supervision, Writing – review & editing, Visualization, Validation. KE: Writing – original draft, Conceptualization, Writing – review & editing, Validation, Supervision. PC: Writing – original draft, Supervision. SE: Writing – review & editing, Validation, Project administration, Methodology, Data curation, Conceptualization, Supervision.

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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.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1341212/full#supplementary-material

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Patient safety culture in private hospitals in China: a cross-sectional study using the revised Hospital Survey on Patient Safety Culture

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Background: This study aimed to translate the revised Hospital Survey on Patient Safety Culture (HSOPSC 2.0) to Mandarin, evaluate its psychometric properties, and apply it to a group of private hospitals in China to identify the determinants associated with patient safety culture.

Methods: A two-phase study was conducted to translate and evaluate the HSOPSC 2.0. A cross-cultural adaptation of the HSOPSC 2.0 was performed in Mandarin and applied in a cross-sectional study in China. This study was conducted among 3,062 respondents from nine private hospitals and 11 clinics across six cities in China. The HSOPSC 2.0 was used to assess patient safety culture. Primary outcomes were measured by the overall patient safety grade and patient safety events reported.

Results: Confirmatory factor analysis results and internal consistency reliability were acceptable for the translated HOSPSC 2.0. The dimension with the highest positive response was "Organizational learning - Continuous improvement" (89%), and the lowest was "Reporting patient safety event" (51%). Nurses and long working time in the hospital were associated with lower assessments of overall patient safety grades. Respondents who had direct contact with patients, had long working times in the hospital, and had long working hours per week reported more patient safety events. A higher level of patient safety culture implies an increased probability of a high overall patient safety grade and the number of patient safety events reported.

Conclusion: The Chinese version of HSOPSC 2.0 is a reliable instrument for measuring patient safety culture in private hospitals in China. Organizational culture is the foundation of patient safety and can promote the development of a positive safety culture in private hospitals in China.

KEYWORDS

patient safety culture, adverse events, HSOPSC 2.0, private hospital, China

1 Introduction

Patient safety is the foremost priority of global health. It is the fundamental requirement for medical care and one of the key domains of quality management at all levels of healthcare (1). Over 1 in 10 patients continue to be harmed from safety lapses during their care. Globally, unsafe care results in well over 3 million deaths each year. In the developing world, as many as 4 in 100 people die from unsafe care (2, 3). One identified reason for such unsafe patient care is a weak safety culture. The shared attitudes, beliefs, and values of all employees lead to behavioral norms in the organization. These norms create a cultural climate of patient safety that promotes consistent performance for patient safety, which is associated with improved patient outcomes (4). As an integral component of healthcare quality management, a positive patient safety culture improves the attitudes and perceptions of patient safety at an individual level, reduces the occurrence of adverse events, and improves the overall safety of the healthcare delivery system (5, 6).

Measurement of patient safety culture in China is limited and has only been conducted in a few public hospitals and in almost no private hospitals (7). Since 2011, China's healthcare reform has emphasized private investment in healthcare services, and driven by domestic demand and policy, the number of private hospitals has risen from 30.8% of the total number of hospitals in China in 2009 to 67.7% in 2021 (8). Private hospitals have gradually become an important part of China's healthcare system to meet diverse healthcare needs.

The Hospital Survey on Patient Safety Culture (HSOPSC), developed by the Agency for Healthcare Research and Quality (AHRQ), is the most widely used instrument to measure safety culture in healthcare organizations internationally. In 2019, AHRQ released a revised version, HSOPSC 2.0, and encouraged its use in place of the original version (9). A previous study contributed to the availability of a Chinese version of the revised surveys on patient safety culture, but it focus on the public hospital nursing team (10). Considering that public and private hospitals setting is very different, and private hospitals constitute a larger proportion of registered hospitals in China currently. Therefore, this study translated the HSOPSC 2.0 to Mandarin and validated it for use in private hospitals in China, also captures a wider breadth of professions instead of a single discipline. The purpose was to identify strengths and areas for improvement related to organizational culture and patient safety by measuring the perception of patient safety culture and identifying the factors associated with overall patient safety grade and adverse events reporting. The study results could provide a reference for managers and policymakers to promote the construction of a patient safety culture in private healthcare institutions.

2 Materials and methods

2.1 Study design and setting

A two-phase practice was conducted to translate and evaluate the HSOPSC 2.0. The original English version was translated into Mandarin and adapted following published recommendations (11). Each item was assessed for clarity and cultural relevance and then evaluated for internal consistency and construct validity. Subsequently, a descriptive cross-sectional study was conducted using the bilingual version.

This cross-sectional study was conducted in nine private hospitals and 11 clinics across six cities in China from 7 to 28 February 2022. The sampling method used in this study was convenience sampling; all the hospitals and clinics are within a group network. An online survey was used to avoid personal contact due to the Coronavirus disease 2019 (COVID-19) pandemic. The survey was strictly anonymous to ensure the privacy of the respondents. All participants were informed about the intention of the study. Two blind researchers were previously instructed about the questionnaire's content and provided with specific training on quality control. They addressed questions and clarified doubts if participants did not understand the questionnaire.

Efforts were made to reduce nonresponse rates and mitigate potential biases introduced by non-respondents. Reminders through emails and instant messaging software were send to participants on a weekly basis, and a lucky draw was conducted at the end, encouraging completion of the questionnaire. Furthermore, the survey was designed to be strictly anonymous, to ensure participant privacy and reduce social desirability bias. Participants were assured that their responses would remain confidential, which encouraged more honest and representative participation.

Ethical clearance and approval was obtained from the Ethics Committee of Beijing United Family Hospital. Considering that this survey is purely anonymous and does not involve the personal information of participants, the committee approved that participants do not need to sign the informed consent and can be informed the intent of the study at the beginning of the questionnaire.

2.2 Instrument

Patient safety culture is conceptually complex and can be viewed within the Patient Safety Culture Theoretical Framework, which is made up of these components: (a) degree of psychological safety, (b) degree of organizational culture, (c) quality of culture of safety, (d) degree of high reliability organization, (e) degree of deference to expertise, and (f) extent of resilience (12).

To measure this complex concept, one commonly used instrument is the Agency for Healthcare Research and Quality Hospital Survey on Patient Safety Culture. The HSOPSC 2.0 questionnaire was developed in 2019 and used in this study to assess hospital staff's perceptions of patient safety culture. The latest version, 2.0, reduced the number of survey items from 51 to 40 and dropped the dimensions from 12 to 10. Some items were reworded because they were sensitive, semantically redundant, or difficult to translate (11). "Does not apply/Do not know" response option was added to each item. Meanwhile, HSOPSC 2.0 changed the response options from 'failing' to 'poor' and 'acceptable' to 'good' in overall patient safety grade.

The questionnaire was translated into Mandarin and modified to fit the Chinese hospital setting. Two independent translators did the preliminary English-to-Chinese translation. Each preliminary version was blindly back-translated by two other people. All

Abbreviations: AHRQ, Agency for Healthcare Research and Quality; CFA, Confirmatory factor analysis; HSOPSC, Hospital Survey on Patient Safety Culture; KMO, Kaiser-Meyer-Olkin; OECD, Organization for Economic Co-operation and Development.

translators were bilingual and with expertise in health, including employees at the World Health Organization, physicians in tertiary hospitals, and students at the University of Virginia School of Medicine. The preliminary version was then evaluated by a translation committee of five bilingual medical professors and synthesized into the latest version.

2.3 Participants and data collection

This study recruited 3,881 hospital staff, including physicians, nurses, technicians, and administrators. A total of 3,064 participants completed the online survey, giving a response rate of 78.95%. The responses were examined for incomplete or invalid data. The exclusion criteria were as follows: (1) surveys were completely blank; (2) contained "Does not apply/Do not know" responses for all survey items; or (3) contained the same answer for all the items. Two invalid questionnaires were eventually excluded, leaving 3,062 questionnaires for analysis.

2.4 Statistical analysis

The internal consistency of the translated HSOPSC 2.0 was assessed using Cronbach's α coefficient, with a value of 0.7 considered acceptable. The appropriateness of the factor analysis was evaluated using the Kaiser-Meyer-Olkin (KMO) test (>0.5) and Bartlett's test of sphericity ($p < 0.05$). Confirmatory factor analysis (CFA) was performed using AMOS version 24 to confirm the factor structure of the questionnaire. As recommended by Jackson (13), we evaluated the following goodness-of-fit indices of the measurement model: Chi-square goodness of fit ($p > 0.05$), root mean square error of approximation (RMSEA, < 0.08), standardized root mean residual (SRMR, < 0.05), and comparative fit index (CFI, > 0.9).

Categorical variables were presented as frequencies with percentages. Differences in categorical outcomes were assessed using the Chi-square test. Percentage of positive responses for each item and dimension were calculated. Responses of 'agree' or 'strongly agree' and 'always' or 'most of the time' for the positively worded items indicated positive responses. Additionally, responses of 'disagree' or 'strongly disagree' and 'never' or 'rarely' for the negatively worded items indicated positive responses. Positive response rates were used to evaluate attitudes toward patient safety culture in different dimensions. A positive response rate > 75% indicated a strong area of safety culture, while <50% needed improvement (14).

The relationship between the explanatory variables (demographic characteristics and 10 dimensions of patient safety culture) and the outcome variables (overall patient safety grade and the number of patient safety events reported) was examined using binary logistic regression. The outcome variable was dichotomized into high ('excellent' and 'very good') and low ('poor' to 'good') overall patient safety grades, and 'none' and '1 or more' reported patient safety events. We treated staff position, duration working for the hospital, working hours per week, and contact with patients as dummy variables and used a forward stepwise logistic regression approach. Multicollinearity in the logistic model was checked using the variance inflation factor (VIF < 10). The goodness-of-fit of the models was assessed using the Hosmer-Lemeshow test ($p > 0.05$). All statistics were managed with Office Excel 2010 and analyzed using IBM SPSS version 24.0. Two-sided p-values <0.05 were considered significant.

3 Results

3.1 Characteristics of respondents

A total of 3,062 respondents from nine hospitals and 11 clinics in six cities across China completed the survey. The mean age of the respondents was 41 (SD 10) years old, and most were female (79.5%). The majority of the respondents (72.8%) had at least an undergraduate degree, 523 (17.1%) were physicians, 1,002 (32.7%) were nurses, and 911 (29.8%) were administrative staff. Of the respondents, 39.3% had worked in the hospital for 1–5 years, while 27.12% worked for 6–10 years. Half of respondents (50.2%) worked 30–40 h a week. Additionally, 74.8% of respondents had direct contact with patients.

3.2 Dimensionality, reliability and validity of the instrument

The dimensionality of the translated HSOPSC 2.0 was assessed using confirmatory factor analysis. The results supported the proposed factor structure, indicating that the instrument captured the intended dimensions of patient safety culture in the context of private hospitals in China.

The HSOPSC 2.0 has been widely used and validated in various countries. The Cronbach's α for the 10 subscales ranged from 0.67 to 0.89 in the U.S. study and 0.61 to 0.83 in the Korean study (11, 15), which provides evidence of internal consistency reliability. In this study, the overall Cronbach's α of the HSOPSC 2.0 was 0.91, and Cronbach's α coefficients of the subscales ranged from 0.52 to 0.88, slightly lower than the other study, which still implied acceptable reliability.

Bartlett's test demonstrated a sufficient inter-item correlation ($p < 0.001$), and the KMO test (0.92) indicated a high model adequacy. The Chi-square test was statistically significant: χ^2/df ratio = 8.77 ($\chi^2 = 3664.95$, df = 418, $p < 0.001$), possibly due to the large sample size. The other multiple indices indicated that the ten-factor model provided a good fit to the data: RMSEA = 0.05 (<0.08), SRMR = 0.048 (<0.05), and CFI = 0.91 (> 0.9).

These findings indicate that the adapted survey instrument, the Mandarin version of HSOPSC 2.0, is valid and reliable for measuring patient safety culture in private hospitals in China.

3.3 HSOPSC 2.0 score

Among these 10 safety culture dimensions, seven were strength areas with over 75% positive response rate. The other three dimensions ranged from 51 to 73%. There were no dimensions with a positive response rate below 50%, indicating a need for improvement. In this study, the highest positive response rate dimension was "Organizational learning - Continuous improvement" (89%), and the lowest was "Reporting patient safety event" (51%). Table 1 shows the

TABLE 1 Scores of the HSOPSC 2.0 and each dimension.

Safety culture dimensions	Average positive response rate (%)			
	This study	Rank	2021 U.S.*	Rank
Organizational learning - continuous improvement	89	1	72	5
Teamwork	88	2	82	1
Supervisor, manager, or clinical leader support for patient safety	86	3	80	2
Handoffs and information exchange	83	4	64	8
Hospital management support for patient safety	82	5	67	7
Communication about error	77	6	64	8
Communication openness	77	7	75	3
Communication about error	73	8	71	6
Staffing and work pace	55	9	58	10
Reporting patient safety event	51	10	74	4
Composite measures average	76		71	

*U.S. Positive response rate published by the AHRQ.

average positive response rates of composite measures for this study were higher than the 2021 U.S. database report (16).

Most participants (65%) rated their unit "excellent" (27%) or "very good" (38%) in patient safety, which was slightly lower than the U.S. rate (69%). Less than half of the respondents (44%) reported at least one event in their hospital in the past year, similar to the U.S. report (46%).

3.4 Univariate analysis of factors correlated with patient safety grade and patient safety events reported

As shown in Table 2, staff position, working time in the hospital, and contact with patients contributed to significant differences in the overall patient safety grade and the number of events reported ($p < 0.05$). Moreover, working hours per week led to significant differences in the reported number of events ($p < 0.05$).

3.5 Binary logistic regression analysis for patient safety grade and patient safety events reported

Regarding the overall patient safety grade, the model was well calibrated (Hosmer-Lemeshow test, $p = 0.992$), and there was no multicollinearity problem (all VIF < 10). The binary logistic regression showed that only staff position and working time in the hospital were influencing factors. Nurses and long working time in the hospital were associated with lower assessments of overall patient safety grades. The binary analysis showed that seven dimensions of patient safety culture were significantly associated with overall patient safety grade (Table 3). A higher level of patient safety culture indicates an increased probability of a high overall patient safety grade.

Regarding the number of reported patient safety events, the model is well calibrated (Hosmer-Lemeshow test, $p = 0.916$), and there is no multicollinearity problem (all VIF < 10). The respondents who had contact with patients, long working time in the hospital, and long working hours per week reported more patient safety events. Nurses, administrators, support, and other clinical positions reported more patient safety events than physicians. Furthermore, Table 4 shows that the incidence of patient safety events reported was closely related to higher levels of patient safety culture.

4 Discussion

4.1 Statement of principal findings

Medical practice is a complex domain with high risks, uncertainties, and layered dynamics. Developing a safety culture is a cost-effective strategy for building a safer healthcare system (17). Efforts to promote a safety culture are associated with better patient outcomes, improved efficiency, and fewer adverse events (1). However, few studies address patient safety as a health strategy for strengthening the private hospital system. This study is the first of its kind in China to explore safety culture issues and the influencing factors in private hospitals.

The overall average positive response rate (76%) was higher than studies conducted in South Korean hospitals (43%) and American hospitals (71%), indicating higher levels of patient safety culture among the staff in this study (13, 15). Among the 10 dimensions of patient safety culture, "Organizational learning - Continuous improvement" was the highest contributing dimension for overall patient safety culture. This implies that the sampled hospitals paid more attention to patient safety issues by providing resources to support patient safety matters and making continuous improvements. Recently, more hospitals in China have actively created an organizational atmosphere of learning while working. Creating a culture of learning and sustainable development is linked to both individual capacity building and organizational performance, and it is considered to be an important factor in facilitating safer and more efficient healthcare delivery (18). Furthermore, "Teamwork" has emerged as one of the top two highest positive response rate dimensions in almost all HSOPC studies, especially in China (19, 20). Previous studies also found that Chinese have a greater appreciation for collectivism (21). This might be associated with Chinese tradition,

TABLE 2 Univariate analysis results.

	Overall patient safety grade (*N* = 3,062)		Patient safety events reported (*N* = 3,062)			
	High	Low	*p* value	None	1 or more	*p* value
Region			0.763			0.731
North China	1,033(52.0)	560(52.1)		884(51.8)	709(52.3)	
East China	750(37.7)	412(38.4)		656(38.5)	506(37.3)	
South China	205(10.3)	102(9.5)		166(9.7)	141(10.4)	
Staff position			<0.001			<0.001
Physicians	333(16.8)	190(17.7)		295(17.3)	228(16.8)	
Nurses	563(28.3)	439(40.9)		493(28.9)	509(37.5)	
Administrator	647(32.5)	264(24.6)		554(32.5)	357(26.3)	
Support	231(11.6)	95(8.8)		219(12.8)	107(7.9)	
Other clinical position	197(9.9)	80(7.4)		125(7.3)	152(11.2)	
Others	17(0.9)	6(0.6)		20(1.2)	3(0.2)	
Working time in the hospital (year)			<0.001			<0.001
< 1	486(24.4)	193(18.0)		504(29.5)	175(12.9)	
1–5	802(40.3)	400(37.2)		643(37.7)	559(41.2)	
6–10	483(24.3)	347(32.3)		390(22.9)	440(32.4)	
≥ 11	217(10.9)	134(12.5)		169(9.9)	182(13.4)	
Working hours per week (hour)			0.554			<0.001
< 30	52(2.6)	23(2.1)		54(3.2)	21(1.5)	
30–40	1,005(50.6)	531(49.4)		888(52.1)	648(47.8)	
> 40	931(46.8)	520(48.4)		764(44.8)	687(50.7)	
Contact with patients			<0.001			<0.001
Yes	1,446(72.7)	843(78.5)		1,170(68.6)	1,119(82.5)	
No	542(27.3)	231(21.5)		536(31.4)	237(17.5)	

which encourages collectivist theories and places relatively more emphasis on cooperation.

4.2 Interpretation within the context of the wider literature

This study demonstrated that the areas with the most potential for improvement were "Staffing and work pace" and "Reporting patient safety event." The relatively low positive response rate for "Staffing and work pace" is similar to previous studies in South Korea (15). According to the Organization for Economic Co-operation and Development (OECD), there were 3.1 nurses and 2.2 doctors per 1,000 population in China, 7.9 nurses and 2.5 doctors per 1,000 population in Korea, which were both well below the average level of about 8.8 nurses and 3.6 doctors (22). Chinese medical staff work in a challenging environment with staff shortages and heavy workloads, which could potentially contribute to clinician burnout and increase the risk of patient safety events (23). One possible explanation is that health workers face a greater workload and intensity because of the COVID-19 pandemic. One of the continuing negative effects of the increase in patient numbers and care intensity is staff shortages. Therefore, the government should take measures to increase the number of medical staff and rationally plan and allocate medical resources.

From 2007 to 2022, seven versions of the Patient Safety Goal were released by the Chinese Hospital Association, each of which included the goal of encouraging medical staff to report patient safety events voluntarily (7). Although there is a consensus that physicians are important in patient safety, this study found that they were less likely to report events, consistent with previous findings in China (24). Compared with physicians, nurses spend more time communicating with patients, giving them more opportunities to identify and report patient safety concerns. Likewise, administrators reported more events, possibly because they placed more emphasis on patient safety or had easier access to the reporting system. A surprising finding of this study is that longer years of service were associated with higher reporting of patient safety events and, by contrast, lower overall perception of safety grades. A possible explanation is that as seniority increases, the experiences, social interactions, perceptions, and values related to patient safety become more complex. Senior medical staff resuscitate acute critical patients, are exposed to higher medical risks, are more aware of the safety practices and benefits of reporting conducted within the hospital, and are more concerned about patient safety (25). This indicates that a culture of improvement is as important to patient safety as a culture of reporting.

TABLE 3 Binary logistic regression models with overall patient safety grade.

	OR(95%CI)	*p* value
Staff position		
Physicians (reference)		
Nurses	1.27(1.00–1.62)	0.052
Administrator	0.61(0.47–0.79)	<0.001
Support	0.49(0.35–0.69)	<0.001
Other clinical position	0.66(0.46–0.93)	0.017
Others	0.40(0.14–1.17)	0.093
Working time in this hospital (years)		
< 1 (reference)		
1–5	1.27(1.01–1.59)	0.039
6–10	1.86(1.46–2.37)	<0.001
≥ 11	1.73(1.27–2.35)	<0.001
Teamwork	0.74(0.64–0.86)	<0.001
Staffing and work pace	0.87(0.81–0.94)	<0.001
Response to error	0.86(0.80–0.94)	0.001
Supervisor, manager, or clinical leader support for patient safety	0.87(0.77–0.99)	0.027
Communication openness	0.83(0.78–0.90)	<0.001
Reporting patient safety event	0.85(0.76–0.94)	0.002
Hospital management support for patient safety	0.68(0.61–0.75)	<0.001

TABLE 4 Binary logistic regression models with patient safety events reported.

	OR(95%CI)	*p* value
Region		
North China (reference)		
East China	1.07(0.90–1.26)	0.459
South China	1.47(1.12–1.93)	0.006
Staff position		
Physicians (reference)		
Nurses	1.35(1.07–1.70)	0.011
Administrators	1.13(0.88–1.45)	0.330
Support	1.47(1.03–2.09)	0.032
Other clinical position	1.87(1.36–2.57)	<0.001
Others	0.34(0.09–1.21)	0.095
Working time in the hospital (year)		
< 1 (reference)		
1–5	2.42(1.95–3.01)	<0.001
6–10	3.14(2.49–3.97)	<0.001
≥ 11	2.95(2.21–3.95)	<0.001
Working hours per week (hour)		
< 30	0.53(0.31–0.92)	0.024
30–40	0.78(0.67–0.92)	0.002
> 40 (reference)		
Contact with patients		
Yes	1.76(1.41–2.20)	<0.001
No (reference)		
Communication about error	1.12(1.01–1.23)	0.029
Communication openness	1.08(1.01–1.15)	0.036
Reporting patient safety event	1.47(1.33–1.62)	<0.001
Hospital management support for patient safety	0.79(0.71–0.87)	<0.001
Handoffs and information exchange	1.14(1.05–1.24)	0.001

Chegini et al. (26) demonstrated that the number of events reported in private hospitals was higher than in public hospitals, which may be the different safety-relevant interventions related to reporting, analysis, and prevention of adverse events in the public and private sectors. Private hospitals are more concerned with a patient-safety-oriented management approach to improve the quality and safety of care (27). In the univariate analysis, employees with less than 1 year of service reported fewer patient safety events than employees with 1–5 years and 6–10 years of service. This may be linked to organizational culture, where individuals' perceptions converge as their time in the organization increases. Thus, organizational culture is fundamental to patient safety.

4.3 Strengths and limitations

This study has several strengths, including that it is the first study in China to explore patient safety culture issues and its influencing factors in private hospitals. Additionally, HSOPSC 2.0 was translated and tested for application in China, which provides a reference for safety culture assessment. Moreover, some influencing factors related to patient safety culture were identified. Nevertheless, some limitations should be considered in this study despite its strengths. The Cronbach's α in some dimensions was lower than 0.7, which might be due to cultural differences between China and the U.S. Furthermore, the generalizability of the findings is not sufficiently clear due to the convenience method, the potential for nonresponse bias in the online survey, and the absence of thorough discussion on confounding factors. However, the response rate was actually high so largely mitigated. Moreover, data for this study were collected exclusively from private hospitals, and all sampled hospitals are part of a large healthcare organization, self-reported data may introduce biases. Further research should be undertaken to extend the scope and sample size as well as compare public and other private hospitals in China.

4.4 Implications for policy, practice, and research

It is important to acknowledge that patient safety depends on a systems approach, which requires contributions and collaboration from various stakeholders. The results of this study suggest that hospitals and healthcare organizations should have imperatives to (1) establish a non-punitive, high-security, and voluntary reporting culture; (2) rationally allocate human resources and work intensity to focus on insecurity and dysphoria among nurses and medical staff; (3)

establish a culture of improvement to promote positive feedback on reporting; and (4) integrate patient safety education into teaching curriculum and clinical practice to establish an organizational culture. These will be important strategies with far-reaching applicability in ensuring quality care and patient safety.

5 Conclusion

As far as the authors are aware, this is the first study conducted in China to validate HSOPSC 2.0 and evaluate patient safety culture in private hospitals. Developing and maintaining a positive patient safety culture among healthcare staff is widely acknowledged as crucial to improving patient safety in healthcare organizations. HSOPSC 2.0 had satisfactory reliability and validity to be applied in private hospitals in China. Organizational culture can promote patient safety and facilitate the development of a positive safety culture in private hospitals in China. The results of this study provide some evidence for developing effective strategies to promote safety culture to ensure patient safety and quality of care.

Data availability statement

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

Author contributions

YL: Writing – review & editing, Writing – original draft, Validation, Supervision, Software, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. JX: Writing – original draft, Validation, Software, Methodology, Formal analysis, Data curation. XY: Writing – original draft, Resources, Methodology, Investigation, Data curation. LY: Writing – original draft, Validation, Resources, Project administration, Methodology. GL: Writing – review & editing, Supervision, Resources, Methodology, Conceptualization. AM: Writing – review & editing, Visualization, Supervision, Resources, Project administration, Methodology, Conceptualization.

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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.

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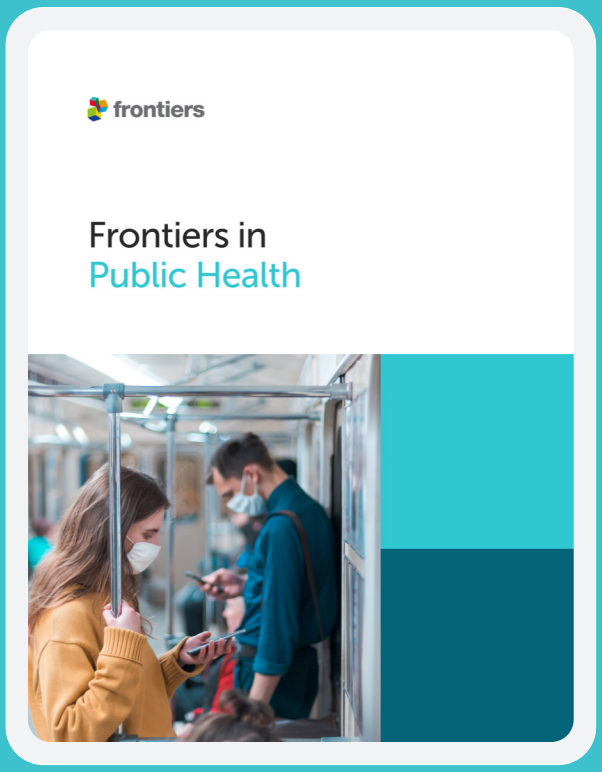

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