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The epidemiology of missed and delayed medical diagnosis: implications for health equity and public health

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The epidemiology of missed and delayed medical diagnosis: implications for health equity and public health

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Editorial: The epidemiology of missed and delayed medical diagnosis: implications for health equity and public health

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Editorial on the Research Topic
The epidemiology of missed and delayed medical diagnosis: implications for health equity and public health

Overview of the Research Topic

The pursuit of diagnostic excellence and the reduction of diagnostic errors improve patient safety and public health. However, the broad range of factors leading to missed or delayed diagnoses at any point in time complicates the identification of and interventions on modifiable factors that reduce or prevent diagnostic errors. Although the importance of diagnostic safety and the costs in terms of human health as well as medical care expenditures are appreciated, the level of research effort and investment into identifying preventable causes of diagnostic errors has been lagging. This in part likely stems from deficiencies in defining specific types of diagnostic error and the lack of standard research approaches for identifying root causes and especially preventable ones. Medicine is practiced in an increasingly complex socio-technical system where causal relationships between system attributes and outcomes including accurate diagnosis may not be visible using traditional epidemiological methods. Additionally, the stigma surrounding missed, delayed or wrong diagnosis no doubt intimidates some practitioners, and subsequently impedes if not precludes the objective examination of all the systemic, institutional and patient factors involved in the science and art of diagnosis. Furthermore, the role of the patient in quality diagnosis is increasingly acknowledged, but how effective patient participation can be enhanced across diverse age, gender, cultural, educational and socio-economic groups remains unclear and challenging.

The epidemiology of missed and delayed diagnosis: implications for health equity and public health, is a collection of invited papers with a focus on epidemiological approaches and perspectives in improving methods leading to an understanding of the preventable causes of diagnostic error, including health equity and public health aspects. This provided a forum in which contributions from various stakeholders were intended to stimulate the exchange of ideas and scientific approaches that transcend professional niches to inspire additional

epidemiological research on diagnostic excellence. As expected, the 10 published articles represent a mix of topics, study approaches, and professional perspectives that share a central theme of improving diagnoses.

Hunter et al. set the epidemiological methods stage by highlighting the challenges but also the criticality of defining suboptimal diagnoses, preferably in ways that objectively can be measured and evaluated against a range of possible risk factors from many different domains (and not simply practitioner training or performance). They discuss how diagnostic errors likely arise due to multiple coincidental "partitioning factors" including abstract factors reflecting individual behaviors, beliefs and communication barriers. They conclude that guided by well-constructed research hypotheses, critical thinking and adherence to good epidemiological methods, insights into contributors to diagnostic excellence will be identified.

Five papers, though addressing different research topics, report on original research conducted to elucidate different aspects of diagnostic error.

McDonald et al. examined data from a representative sample of US patient survey responses regarding their diagnostic experiences. Over one third of the sample reported experiencing a "diagnostic problem or mistake" in the preceding 4 years. The subgroup analyses and reported associations raise interesting questions for promising future study.

Maleki et al. explored sociodemographic inequalities in the postnatal care coverage (PNC) provided women in Iran. They noted that these disparities and the failure to deliver proper PNC to all women regardless of age educational level, region, etc., increases the risk of adverse postnatal health consequences.

During the COVID-19 experience in China, Wang et al. demonstrated that self-care practices resulting from limited access to care providers was common, although certain subgroups of the population were less inclined to maintain regular exercise and weight control routines.

Another study in China compared health self-assessment ratings of those enrolled in the Urban and Rural Residents Medical Insurance (URRMI) with those not enrolled. Yu et al. reported that those enrolled in the URRMI reported more favorable health self-assessments.

Atac et al. presented an interesting study in which family physicians estimated the probability of diagnosis in three clinical scenarios about cancers (breast, cervical, and colorectal) and three infections (pneumonia, urinary tract infection, and COVID-19). For all scenarios, physicians' estimates were higher than the evidence range.

One paper described a protocol for a planned study of improving communication about diagnosis in pediatric care. Rasooly et al. frame pediatric diagnosis as a process stemming from "systems-of-work" communication and propose methos for assessing the validity of various diagnostic error detection methods.

Syros et al. present findings from a systematic review of the literature on barriers to care experienced by musculoskeletal sarcoma patients. They defined four types of barriers to obtaining appropriate care, including socioeconomic, geographic, healthcare quality and sociocultural factors, noting that assessing these can lead to beneficial interventions to improve quality and reducing delays in obtaining care.

Two commentaries rounded out the Research Topic. Coronado-Vázquez et al. describe a model for supporting cancer prevention and early diagnosis and treatment of cancer among adults experiencing homelessness in Madrid, Athens, Vienna, and Cambridge. They concluded that the structural injustices in the health systems in these regions, including recognizing citizenship and simple "generosity" must be addressed to reduce health inequities faced by this population.

Another commentary presented a compelling argument for critically assessing the necessity and problems with using race and ethnicity in diagnostic, treatment and other clinical support tools. Using the Vaginal Birth After Cesaran (VBAC) calculator as a case study, Kimani summarized the use of racial and ethnic categories in science and medicine historically and currently, and concludes that medical algorithms based on these interfere with efforts to reduce maternal morbidity and mortality.

This Research Topic provides a wide range of perspectives and approaches to investigate and understand the important public health problem of preventable harm caused by missed and delayed diagnosis. The articles offer novel insights, methods to emulate, and applications of epidemiological tools to several populations and disease entities. Diagnostic processes and the settings where they occur are rapidly changing with more remote care, patient self-care, artificial intelligence and other advanced diagnostic technology. Future research on these systems and new methods to understand the causal relationships between process and system attributes and diagnostic safety outcomes will require better epidemiological methods that identify areas for improvement, leading to better patient and public health.

Author contributions

DS: Conceptualization, Supervision, Writing – original draft, Writing – review & editing. KM: Conceptualization, Supervision, 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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Family physicians overestimate diagnosis probabilities regardless of the test results

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Introduction: As useful tools for clinical decision-making, diagnostic tests require careful interpretation in order to prevent underdiagnosis, overdiagnosis or misdiagnosis. The aim of this study was to explore primary care practitioners' understanding and interpretation of the probability of disease before and after test results for six common clinical scenarios.

Methods: This cross-sectional study was conducted with 414 family physicians who were working at primary care in Istanbul via face-to-face interviews held between November 2021 and March 2022. The participants were asked to estimate the probability of diagnosis in six clinical scenarios provided to them. Clinical scenarios were about three cancer screening cases (breast, cervical and colorectal), and three infectious disease cases (pneumonia, urinary tract infection, and COVID-19). For each scenario participants estimated the probability of the diagnosis before application of a diagnostic test, after a positive test result, and after a negative test result. Their estimates were compared with the true answers derived from relevant guidelines.

Results: For all scenarios, physicians' estimates were significantly higher than the scientific evidence range. The minimum overestimation was positive test result for COVID-19 and maximum was pre-test case for cervical cancer. In the hypothetical control question for prevalence and test accuracy, physicians estimated disease probability as 95.0% for a positive test result and 5.0% for a negative test result while the correct answers were 2.0 and 0%, respectively ($p < 0.001$).

Discussion: Comparing the scientific evidence, overestimation in all diagnostic scenarios, regardless of if the disease is an acute infection or a cancer, may indicate that the probabilistic approach is not conducted by the family physicians. To prevent inaccurate interpretation of the tests that may lead to incorrect or unnecessary treatments with adverse consequences, evidence-based decision-making capacity must be strengthened.

KEYWORDS

primary care, family medicine, general practice, diagnostic tests, likelihood ratios

1 Introduction

Diagnostic tests are helpful tools to facilitate deciding the correct diagnosis in line with the medical history and symptoms of the patients in terms of clinical decision-making (1). Application of the principles of evidence-based medicine helps clinicians make better diagnostic and management decisions. All diagnostic procedures, including laboratory tests, are based on probability estimations that need careful interpretation. Unnecessary request or misinterpretation of diagnostic tests may lead to underdiagnosis, overdiagnosis or misdiagnosis (2). Misinterpretation can adversely affect treatment, recovery, and health expenditures (3).

The accuracy of the tests which can be estimated as sensitivity, specificity and predictive values is decisive when deciding on diagnosis and treatment. The prevalence of diseases is also a measure which strongly influences the positive predictive value: the lower the prevalence, the lower is the probability of being sick even after a positive test (4).

Methods regarding accuracy, reproducibility and probability estimations for diagnostic tests are provided to medical school students through clinical epidemiology and evidence-based medicine curriculum (5). However, studies show that physicians do not comprehensively understand and interpret the probabilities during their clinical practices (6). The main consequences of this phenomena is overestimation of both positive and negative test results by physicians (7–9). The same problem exists among primary care physicians who have a key role in screening programs and outpatient services (10).

In the Turkish healthcare system primary care and preventive services are provided by family physicians for the registered populations (11). Family physicians are general practitioners (GP) or specialists and have an important role in the screening in breast, cervical and colorectal cancers as a part of national control programs in addition to primary care practices (12). Respiratory system and urinary tract infections are among the most common causes of admission to their offices (13). Therefore, family physicians are expected to use and interpret the test results appropriately and estimate correct probabilities of these diseases. This issue has become more important during the COVID-19 pandemic when tests for infection detection were widely performed, and their correct interpretation was important (14).

In this study, our aim was to explore primary care practitioners' understanding and interpretation of the probability of a disease before and after test results for six common clinical scenarios.

2 Materials and methods

This cross-sectional study was conducted among family physicians working at primary care services in Istanbul during November 2021 and March 2022. Sample size was calculated as 354 assuming a prevalence of risk overestimation of 50%, with 95% confidence intervals within ±5%. All primary care physicians in five geographically dispersed districts of İstanbul (Başakşehir, Eyüpsultan, Fatih, Sultanbeyli, Üsküdar) were included in the study without using any sampling method ($n = 613$). Among them, 414 physicians have participated in the study, with a 67.5% response rate. Data was collected during face-to-face interviews.

The questionnaire consisted of two sections. The first section contained seven questions regarding the participants' sociodemographic and professional characteristics. The second section contained questions about the probability of diseases in six clinical scenarios (Appendix 1). These scenarios are adapted from the study of Morgan et al. (10) in consideration of the common health problems which are expected to be diagnosed and/or treated by family physicians in Türkiye. Scenarios included three cancer types (breast, cervical and colorectal) within scope of the national cancer control program, two frequent infectious diseases (pneumonia and urinary tract infection) in primary care, and ongoing COVID-19 pandemic. Cases in the cancer screening scenarios were asymptomatic while they were symptomatic in infectious disease scenarios. Each scenario was prepared in agreement with the recent literature. A hypothetical control question measuring the understanding of the participants on the accuracy of diagnostic tests was also included.

Mammography for breast cancer, pap smear for cervical cancer, stool occult blood test for colorectal cancer, chest radiography for pneumonia, complete urinalysis for urinary tract infection, and PCR test for COVID-19 were used as diagnostic tests and the participants were asked probability of the given disease in three conditions: (a) before performing the diagnostic test, (b) after a positive test result, and (c) after a negative test result. The participants' responses were compared to the test accuracy values from existing evidence-based literature. True answers to the questions were determined considering the most relevant national and international guidelines for physicians in Türkiye (Appendix 1).

Python programming language was used for data analysis. Descriptive statistics to summarize the data were frequency, percentage, median, interquartile range (IQR) for non-normally distributed continuous variables, the mean, and the standard deviation for normally distributed continuous variables. The Wilcoxon signed-rank test was used to compare physicians' estimates of disease probabilities and probabilities derived from the evidence. Type I error (α) level of 0.05 was used in the interpretation of the significant test results.

This study was conducted with the permission from the Ministry of Health of the Turkish Republic (07/10/2021) and was approved by the Ethics Committee of Non-Invasive Clinical Studies of Istanbul Medipol University (15/10/2021, No: 1021).

3 Results

414 family physicians participated in the study. 53.9% ($n = 223$) of them were male, 86.2% ($n = 357$) general practitioners and the mean age was 42.9 ± 10.3. The characteristics of the study sample are summarized in Table 1.

The density distribution of physicians' estimates of diagnostic probabilities for each scenario is presented in Figure 1. For all scenarios, physicians' estimates were significantly higher than the scientific evidence range (Appendix 2; Supplementary Table S1).

For breast cancer, the median of physicians' estimation was 10.0% (evidence was 0.2%) for pre-test disease probability ($p < 0.001$). Both for cervical and colorectal cancer, the median of physicians' estimation was 5.0% (evidence was, respectively, 0.01 and 0.06%) for pre-test disease probability (for both comparisons, $p < 0.001$). After a positive test result, physicians estimated the disease probability as 50.0% both

TABLE 1 Descriptive characteristics of the participants.

		n	%
District	Üsküdar	104	25.1
	Fatih	101	24.4
	Eyüp	74	17.9
	Başakşehir	74	17.9
	Sultanbeyli	61	14.7
Gender	Male	223	53.9
	Female	187	45.2
	Unknown	4	1.0
Physician group	General practitioner	363	87.7
	Specialist	44	10.6
	Unknown	7	1.7
Total		414	100.0
		n	Mean ± S.D.
Age		410	42.9 ± 10.3
Professional experience (years)		404	16.9 ± 10.5
Experience as GP (years)		370	7.6 ± 4.0
Number of registered populations		405	3373.5 ± 860.2

for breast cancer and cervical cancer while evidence was, respectively, 8.7 and 0.14% (for both comparisons, $p < 0.001$). After a positive test result, physicians estimated the colorectal cancer probability as 40.0% (evidence was 0.74%; $p < 0.001$). When test results were negative, physicians estimated the disease probability as 10.0% (evidence was 0%) for breast cancer, as 5.0% (evidence was 0.0032%) for cervical cancer and 5.0% (evidence was 0.02%) for colorectal cancer (for both comparisons, $p < 0.001$).

For pneumonia and urinary tract infection, physicians' estimations were 20.0% and 40.0%, respectively, for pre-test disease probability, while the evidence was 5.0% and 1.0%. After a positive test result, these estimations increased to 85.0% and 90.0% respectively, while the evidence was less than 10.0%. When the test results were negative, physicians estimated the disease probability as 10.0% whereas the evidence was less than 5.0%.

For COVID-19, the median of physicians' estimation was 80.0% (IQR 50.0–90.0%) for pre-test disease probability while evidence was 56.0% ($p < 0.001$). After a positive test result, physicians estimated the disease probability as 99.0% (IQR 90.0–100.0%) whereas evidence was 95.4% ($p < 0.015$). When test results were negative, physicians estimated the disease probability as 50.0% (IQR 25.0–70.0%) while evidence was 0.04% ($p < 0.001$).

When physicians were provided with prevalence and test accuracy information of the hypothetical control question, they estimated disease probability as 95.0% (IQR 95.0–100.0%) for a positive test while the true answer was 2.0% ($p < 0.001$) and 5.0% (IQR 5.0–10.0%) for a negative test whereas the true answer was 0% ($p < 0.001$).

Table 2 shows the breakdown of disease probability estimations by physician groups. Although specialists showed a slightly lower tendency toward overestimation than general practitioners in several scenarios, their estimates were not close to the evidence ranges. Subgroup analyses showed that female participants estimated probabilities higher than males with a median difference around the range of 2.0–15.0, but estimations of participants were consistent across districts (Appendix 2; Supplementary Tables S2–S3).

Physicians' probability estimations were significantly different from the evidence values (Appendix 2; Supplementary Table S4). Further subgroup analyses by physician group, gender, and district were provided in Appendix 2; Supplementary Tables S5–S7.

4 Discussion

In this study, we investigated diagnostic probability estimations of the family physicians about different scenarios regarding six hypothetical clinical cases. Three of the cases were pneumonia, COVID-19, and UTI, which are frequently encountered infectious diseases in primary care; the other three were cervical cancer, breast cancer, and colorectal cancer, which are routinely screened in primary care within the scope of the national cancer control program. Most striking finding of the study was the overestimation of all diagnoses before and after test results for the given scenarios.

The overestimation in all scenarios shows that the underlying potential causes of overestimation are not specific to the clinical cases but represents a more general problem. It is noteworthy that the overestimation varies between 1.04 times (COVID-19, after positive test) and 1,250 times (cervical cancer, before test), and it is over 10 times in 13 of the 18 alternatives examined. As was found in Morgan et al.'s study, these results are related to the overestimates of pre-test probability (10).

Overestimated responses given to the negative test results in all cases show that physicians overemphasize the symptoms when deciding on the diagnosis. Besides, the overestimated answers given to the hypothetical control question indicate that the physicians do not have comprehensive knowledge of probability estimations. The fact that the overestimation in our study was similar to previous studies reveals the universality of the problem. In a review article investigating how healthcare professionals interpret the results of diagnostic tests, it was stated that the probability estimates were in the direction of overestimating, regardless of whether the test result being positive or negative, and it has been concluded that commonly used measures of test accuracy are poorly understood by health professionals (7).

Family physicians in primary care have a key role in the management of clinical cases examined in our study. They work as individual health consultants who deal with all the health problems of their enrollees, provide preventive services and who are expected to solve the handleable problems at the primary care level, or to refer the complicated, unresolved cases to further levels and then follow up (15). Therefore, their role is important not only in curative services but also in primary and secondary prevention such as cancer screening.

In our study, pretest probability was higher in cancer screening tests than in UTI and pneumonia, whereas it was higher in UTI and pneumonia than in breast cancer screening in Morgan et al.'s study. High levels of overestimation in presented cancer scenarios can be concluded as the general perception of physicians toward cancer screening tests. Comparable results in other studies on this matter have also shown that physicians tend to overestimate the risk of cancer (16, 17).

Probability of UTI was more overestimated than pneumonia in before and after negative test scenarios and this result may be regarded because of physicians' prioritization of the patient's symptoms compared to the test results while diagnosing UTI. According to the

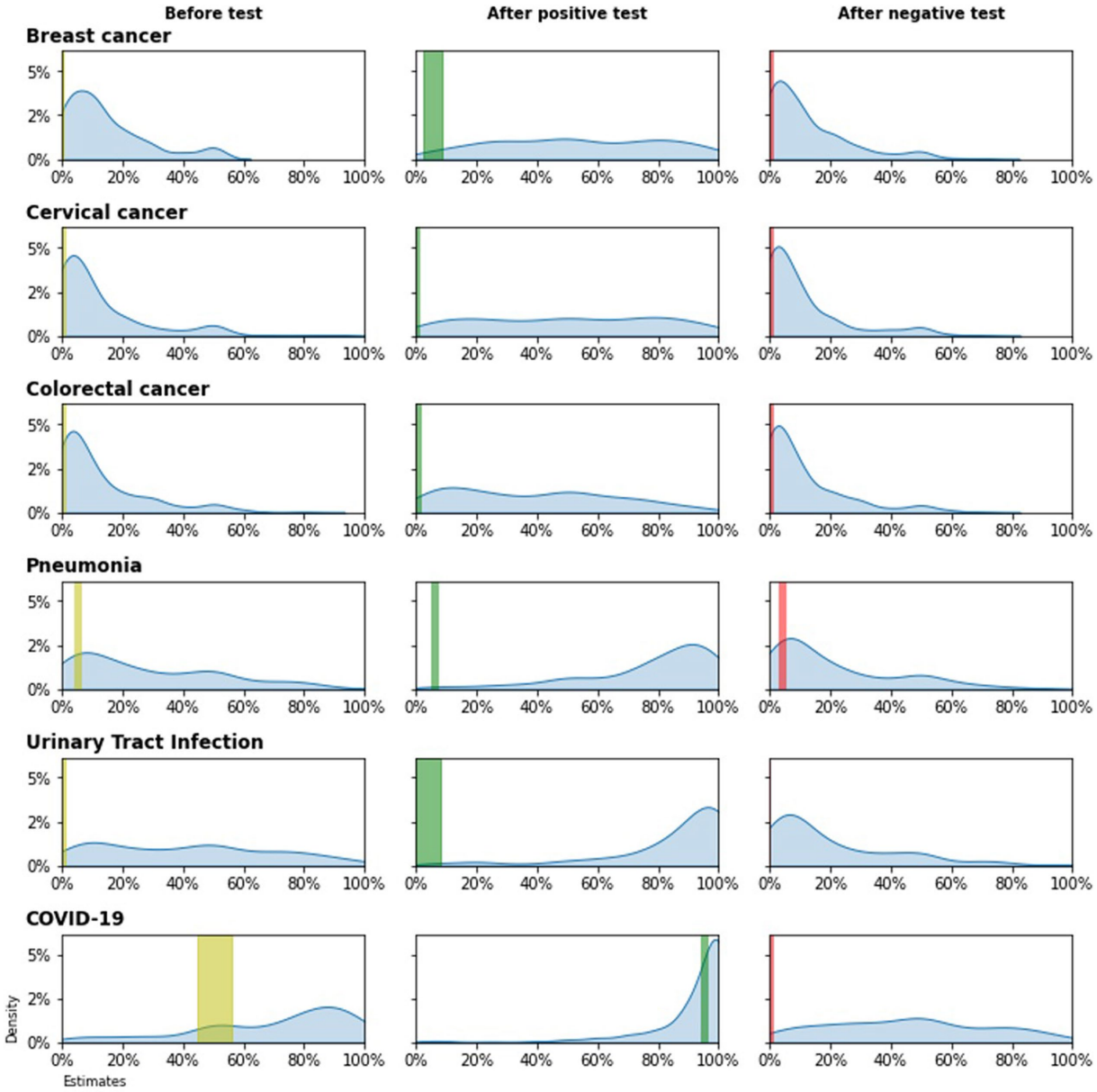

FIGURE 1
Estimates of diagnostic probabilities for each case description* *Colored vertical bars indicate the evidence range.

literature for UTI management, pretest probability, which is estimated with the patient's current symptoms, can significantly affect the post-test probability and in clinically high-risk cases, the post-test probability is evaluated as high even if the test result is negative (18).

There was also an overestimation in COVID-19 case scenarios. However, the frequency of overestimated responses given before test and after positive test result (1.43 and 1.04, respectively) were lower than all other case scenarios. We conclude this finding as the result of availability of up-dated information for COVID-19 management prepared by the Ministry of Health (19).

Diagnostic tests are valuable tools when evaluated together with the patient's symptoms and lead the physician to an exact diagnosis. Although some of them have successful diagnostic performance, some are not the gold standard and only used for screening which requires advanced procedures to confirm diagnosis (17). In fact, diagnosis ultimately depends on the physician's decision. Symptoms, test results and consultations are important parts to decide an appropriate diagnosis and treatment. Current developments in medicine provide test alternatives to physicians, but it is criticized as the dependency on technological solutions puts the experiences of physicians on the shelf and they mostly rely on the test results rather than their own experience (20). From this perspective, while diagnostic tests have a noteworthy role, they may cause overestimation, as seen in our study.

Physicians' decision-making is considered critical for patient safety, as diagnostic errors and inappropriate treatments can harm patients, fail to address actual problems and waste resources (21, 22). It was shown that if family physicians misdiagnose, they mostly misregulate the treatment (23). Misinterpreting test results during

TABLE 2 Estimated disease probabilities for each scenario by physician groups.

Scenarios	Physician Groups, Median (IQR)			Evidence range*, %
	GPs, %	Specialists, %	Both groups, %	
Breast cancer				
Before test	10 (5–20)	10 (4–16)	10 (5–20)	0.2–0.3
After positive test result	50 (30–80)	50 (30–80)	50 (30–80)	2.5–8.7
After negative test result	10 (2–20)	8 (1–20)	10 (2–20)	0
Cervical cancer				
Before test	6 (2–20)	4 (1–10)	5 (2–20)	0.01
After positive test result	50 (22–80)	50 (20–72)	50 (20–80)	0.14
After negative test result	5 (1–15)	3 (1–10)	5 (1–13)	0.0032
Colorectal cancer				
Before test	7 (2–20)	4 (2–10)	5 (2–20)	0.06
After positive test result	40 (15–60)	25 (10–50)	40 (15–60)	0.74
After negative test result	5 (1–15)	4 (1–10)	5 (1–15)	0.02
Pneumonia				
Before test	20 (10–50)	20 (10–50)	20 (10–50)	5
After positive test result	85 (65–90)	80 (75–90)	85 (70–90)	6.2
After negative test result	10 (5–30)	20 (9–38)	10 (5–30)	4.1
UTI				
Before test	45 (20–70)	20 (10–50)	40 (15–65)	0–1
After positive test result	90 (80–100)	90 (75–100)	90 (80–100)	0–8.3
After negative test result	10 (5–35)	10 (2–28)	10 (5–30)	0–0.11
COVID-19				
Before test	80 (50–90)	80 (55–88)	80 (50–90)	45–56
After positive test result	99 (90–100)	96 (90–100)	99 (90–100)	95.41
After negative test result	50 (25–70)	50 (30–68)	50 (25–70)	0.04
Control question				
After positive test result	95 (90–100)	95 (95–100)	95 (95–100)	2
After negative test result	5 (5–10)	5 (5–5)	5 (5–10)	0

UTI, Urinary Tract Infections; IQR, Interquartile range. *See Appendix 1 for the references to the evidence range derived through review of the literature.

decision-making can have adverse effects on the patients. For instance, in cases such as pneumonia and UTI that may require antibiotic treatment, the overestimation can lead to unfavorable outcomes in terms of antibiotic resistance (24).

We found another important overestimation in cancer screening tests. These tests are not definitive diagnostic tools, and they only lead to further procedures to confirm diagnosis. In cancer screenings, it can be interpreted as favorable that physicians attribute more value to screening tests in order not to miss probable cases. However, the risk of labeling for false positive individuals and its social and psychological consequences should always be kept in mind (25).

Post-test probability is expected to be estimated by considering pre-test probability (i.e., prevalence) and test accuracy (i.e., sensitivity and specificity). The reasons for physicians' poor performance in probability estimation are due to lack of knowledge or misunderstanding (26). They mostly do not use likelihood ratio and pre-test probability in their estimations, eventually, test results have been overestimated (7, 27). In a study evaluating physicians' probabilistic approach to the test results, no significant difference was found according to the type of data (sensitivity-specificity vs. likelihood ratios) (28). In another study investigating the change between the pre-test and post-test estimations according to the type of data shared, the successful estimations were 8% for sensitivity-specificity, 34% for the likelihood ratio, and 73% for the graphic form. Researchers have emphasized that clinicians may have difficulties understanding values that require arithmetic calculation, but it is easier for them to understand with visual tools such as graphs (29). Although we did not inquire about the reasons in our study, the poor performance in the hypothetical question may have been caused by a misunderstanding of the question due to the technical terms. An accurate estimation of the probability of the test results is a fundamental competence during clinical decision-making (30), and the lack of knowledge on methodological topics can be eliminated with evidence-based medicine training (31).

When the sensitivity and specificity are constant, the predictive values change according to the prevalence while evaluating the test results (32). However, most physicians cannot consider the prevalence when estimating the probability of disease after a positive test result,

regardless of clinical experience and the institution they work for (26, 29). On the other hand, it is necessary to know the prevalence of the disease in a country or region in order to estimate probabilities correctly during a clinical decision, and ignorance of the frequency is considered a bias (20). Yet, the updated prevalence of major diseases usually is not available for the participants of our study. They should be shared and updated regularly by the health authorities.

Our study has some limitations. Scenarios prepared in line with the current scientific guidelines were directed to physicians. However, we do not know the real situation as cases applying in real world experiences may not fit these ideal scenarios. A limitation is that the diagnostic criteria change in time and there is not any guide specific to the population in which the study was conducted. Except for the COVID-19, international data could be used for probability estimations in scenarios instead of national ones. Because the data were collected as face-to-face observation, the participants may have stated differently from the Hawthorne effect compared to their daily practice. Despite these limitations, the overestimation was excessive in all clinical cases, and cannot only be explained by confounding variables or misinterpretation of the questionnaire.

In conclusion, in the present study, primary care physicians consistently overestimated the actual risk of disease regardless of the results of diagnostic or screening tests, despite the relatively high frequency of such diagnoses, and the availability of well performing tests with their performance parameters. This problem makes it difficult to fulfill patients' needs as it reduces the accurate decision-making by physicians when selecting diagnosis and treatment. The findings of this study indicate the extensiveness and the magnitude of the problem and warrant interventions to improve the quality of primary care. However, to determine the kind of intervention that addresses the issue best, there is a need for qualitative studies to illuminate why and how physicians overestimate the disease probabilities.

Data availability statement

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

Ethics statement

This study involving humans were approved by Ethics Committee of Non-Invasive Clinical Studies of Istanbul Medipol University (15/10/2021, No: 1021). This study was conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

ÖA, HK, AF, AP, SÇ, MNA, MA, MS, YT, and OH: conception and design. MNA, MA, MS, and YT: acquisition of data. ÖA and HK: analysis. ÖA, HK, AF, AP, and SÇ: interpretation. ÖA, HK, AF, AP, SÇ, and OH: draft and revision.

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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Cancer prevention in people experiencing homelessness: ethical considerations and experiences from the CANCERLESS project

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The incidence of cancer in Europe has been increasing in recent years. Despite this, cancer prevention has remained a low priority in health policies. Cancer is one of the main causes of mortality among people experiencing homelessness, who continue to have difficulties accessing prevention programs. A strategy that has been tested to favor cancer prevention is the health navigator figure. The objective of *CANCERLESS* project is to implement this model among populations experiencing homelessness in four European countries to foster the prevention and early detection of cancer. In this perspective, a presentation of *CANCERLESS* project is made, and its ethical aspects are discussed according to the ethics of public health, the ethics of care, solidarity, relational autonomy, and the social recognition of the virtue of just generosity. The ethical foundations of *CANCERLESS* project are rooted in social justice and in equity in access to health systems in general and cancer screening programs in particular. The ethics of public health guided by utilitarianism are insufficient in serving the interests of the most disadvantaged groups of the population. Hence, it is necessary to resort to relational bioethics that includes the ethics of care and solidarity and that recognizes the moral identity of socially excluded persons, reaffirming their position of equality in society. Relational autonomy therefore provides a broader conception by including the influence of living conditions in decisions. For this reason, the *CANCERLESS* project opts for a dialogue with those affected to incorporate their preferences and values into decisions about cancer prevention.

KEYWORDS

homelessness, vulnerability, cancer prevention, autonomy, equity, public health, ethics, care

1 Introduction

According to WHO data, cancer is responsible for 16% of deaths that occur worldwide each year (1).

The incidence of cancer in Europe has been increasing in recent years. A 2018 report that analyzed the incidence of 25 types of cancers in 40 European countries concluded that the number of new cases during that year had been 3.91 million (2). Due to the aging and growth of the population, these are expected to increase in Europe to 4.75 million, with an expected mortality of 32% (3).

Despite this, cancer prevention has remained a low priority for governments and even for WHO (4). Investments have been directed to the development of new treatments, which are much more expensive and not accessible to all, instead of to the promotion of preventive measures with proven benefits (5). We cannot make a point to suggest that an acute disease is less priority, but to make sure we do not forget of the ongoing issues, as well as the effects that infectious disease pandemics will have on the health systems and chronic disease care.

Primary and secondary prevention can reduce the economic and personal costs of cancer, preventing physical and psychological problems as well as those derived from treatments (5).

Homelessness is a very common public health problem in middle- and high-income countries. Just in the European Union, it is estimated that 4.1 million people experience homelessness annually, although its real prevalence is difficult to establish due to the lack of a unified concept of "people experiencing homelessness" and to the different methodologies used to calculate this population, rendering comparability across countries difficult (6). In general, in European countries, a "person experiencing homelessness" is defined as someone who sleeps outdoors or in a shelter (7). European Typology of Homelessness and Housing Exclusion (ETHOS) classifies living situations that constitute homelessness or housing exclusion. ETHOS identifies four main categories of living situation: Rooflessness, Houselessness, Insecure Housing, and Inadequate Housing (8).

The profiles of these populations have changed over time. Currently, young people, women and migrant families are the most affected by homelessness (6).

The physical and mental health of individuals experiencing homelessness are worse than that of the general population (9). A lack of financial resources, substance abuse issues exposure to infectious diseases, increased injuries and reduced access to health services contribute to this statistic (10). Psychiatric morbidity is high, with alcohol and drug use disorders, schizophrenia and depression being the most frequent problems (11). Mortality is also higher, mainly due to infections, ischemic heart disease, substance abuse and injuries (12). It is estimated that, on average, the age at which people experiencing homelessness die is approximately 30 years below that of the rest of the population (13).

Cancer has been described as one of the main causes of mortality in people experiencing homelessness (14), with lung cancer being the most frequent in men associated with high prevalence of tobacco smoking and cervical cancer in women due to associated risk factors including limited knowledge of risk factors, limited access to preventive services and tobacco smoking (15, 16). Among these patients, survival at 10 years is also lower (16), in part due to their usually late diagnosis of cancer owing their difficulty accessing health services.

At times, such access is limited by legal problems or discrimination due to one's homeless status (17). The incidence of advanced cancer is higher among structurally vulnerable populations due to delays in diagnosis and treatment (18). Their structural vulnerability results from poverty, unstable housing, and discrimination based on race and gender (19). In relation to cancer, socioeconomic disadvantages predispose patients to poor medical care (20).

One of the strategies that has been tested to favor the entry of these communities, which experience social exclusion due to homelessness, to health services is the patient navigator model, by whom information is given to people about healthy lifestyles, diagnostic tests and treatments, facilitating their inclusion in screening programs (21). It is a person-centered community health model that has been shown to be effective in improving health outcomes through improved accessibility to health services (22, 23).

The objective of this perspective article is to discuss the ethical aspects of cancer prevention among people experiencing homelessness based on an analysis of the European *CANCERLESS* project.

2 Methodology

2.1 The CANCERLESS project

First, we described the European *CANCERLESS* project, its objectives, participants, design, intervention, and its applicability in clinical practice.

CANCERLESS stands for "Cancer prevention and early detection among the homeless population in Europe: Coadapting and implementing the health navigator model."

The *CANCERLESS* project has three objectives: (1) to develop person-centered health services that promote the access of people experiencing homelessness to cancer prevention and screening; (2) to implement the health navigator model among individuals experiencing homelessness in order to reduce the burden of cancer and associated costs; and (3) to use the knowledge gained for the transformation of cancer care and the implementation of the health navigator model in Europe.

This study was carried out through a longitudinal cohort of people experiencing homelessness from Madrid, Athens, Vienna, and Cambridge. Participants of the *CANCERLESS* project are people aged 18 or over users of homelessness services, who fall under one of the ETHOS categories and who provide their consent to participate.

In each country, the project is expected to recruit an average of 300 people aiming to measure the effectiveness of the focal intervention before, after 4–6 weeks and at end of intervention.

The intervention consists of the implementation of the health navigator model to bring primary and secondary cancer prevention services closer to social care points and facilitate access to the health system for screening. The health navigator model is a combination of the patient navigator model and the patient empowerment model. It consists of different phases: (1) sociohealth assessment of people and determination of biopsychosocial risk; (2) health education through recommendations for cancer risks and screening methods; (3) facilitation of adherence to the screening program through the use of relational techniques that create and reinforce trust between people experiencing homelessness and professionals; (4) agree to and accompany to appointments for screening, coordinating these with

participants, social service centers, and health centers; (5) accompanying patients throughout the entire process until obtaining results through social support; (6) agreeing with the salient professionals to obtain results and negative news reports; (7) follow-up to guarantee care after screening; and (8) to produce agreements with community organizations for greater flexibility in services and/or to generate facilitation channels adapted to people experiencing homelessness.

In the analysis, quantitative and qualitative methods will be combined amid comparisons of different interventions and countries. Health status will be determined with data related to acute and chronic diseases, time of diagnosis, previous participation in cancer screening campaigns, use of health resources, risk behaviors, alcohol and drug use, diagnosis of depression and anxiety [Depression, Anxiety and Stress Scale (DASS)/Brief Symptom Inventory (BSI), self-perceived health (SF-12 Health Survey) and general health status (Short Form of the Self-Administered Multidimensional Prognostic Index, SELFY-MPI-SF) and Cumulative Illness Rating Scale (CIRS)].

In addition, qualitative data will be collected through a quasiexperimental analysis as follows: (1) Determine the causal relationships between "exposure" and "response" (pre-post) to define the causal relationships obtained from the bidirectional analysis of social barrier-determinant impact; (2) Development of facilitators and/or elimination of barriers associated with social determinants on codified navigation actions; (3) Define the adherence rate that allows us to measure the type of performance and time of the professional with regards to the relational objectives; and (4) Delineate the requisite professional profiles and types of skill difficulty concerning adherence.

A cost-effectiveness analysis will be carried out using Monitoring and Assessment Framework for the European Innovation Partnership on Active and Healthy Aging (MAFEIP), which calculates impact indicators such as the incremental value related to age and estimates the accumulated utility based on the cost of innovation and on the improvement in quality of life related to health.

Sociodemographic characteristics and health outcomes will be evaluated across the global sample and separately in each of the countries. The main effects during follow-up will be measured among the total population using an intention-to-treat analysis.

2.2 Literature review

Secondly, in order to discuss the ethical aspects of cancer prevention in people experiencing homelessness, a narrative review of the available studies published in PubMed, Web of Science and Scopus was conducted.

The Medical Subject Headings (MESH) terms included were: Homelessness, Prevention, Cancer, and Ethic.

As eligibility criteria, we defined the inclusion and exclusion criteria based on the population and conceptual framework of the study:

- Population: People experiencing homelessness including those individuals living in supportive housing, transitional housing, unstable housing, inadequate housing, and inappropriate housing.
- Conceptual framework: Access to cancer prevention programs (detection of specific types of cancer, such as breast, cervix, and colon).
- Articles included: Studies conducted in any environment/country/health system. No limitations in terms of sex and gender. Original research and reviews (qualitative, quantitative, and mixed method study designs). Gray literature. Articles published until June 2023 were included.
- Exclusion criteria: Any publication other than original research or review. Not having access to the full text of the publication.

3 Discussion

The *CANCERLESS* project has been designed based on the hypothesis that a health navigator can improve the access of people experiencing homelessness to cancer prevention and screening programs, acting as a link between this population and health services and thereby overcoming the barriers that these systems interpose.

The following questions are therefore raised: (1) according to the ethics of public health, the focus should be placed on minority populations who are excluded from cancer prevention and screening programs due to the determinants that they present as indicators of social exclusion; (2) it is necessary to resort to the principle of solidarity when designing public health policies for cancer prevention; and (3) autonomy (liberal) is insufficient in its application among people experiencing homelessness. We must resort to relational autonomy, which has a broader vision of the influence of living conditions on decision-making, as well as social recognition and the virtue of just generosity, which respect citizenship and expand the vision of the obligations of the State to achieve it, thereby preventing the social abandonment suffered by people experiencing homelessness.

3.1 Ethics of public health and ethics of care

Prevention measures in public health originated in consequentialism, whereby actions are justified based on their consequences and utility. In terms of cancer prevention campaigns, public policies are directed to the benefit of the majority of the population; minority and excluded groups, those who have difficulties accessing health services, are often discarded from these proceedings.

To counteract excessive consequentialism, the integration of virtues into public health decision-making has been proposed (24). According with the ethics of virtues, health policies would be enriched by introducing the perspectives of different kinds of people.

Among the principles of the ethical practice of public health is the recognition of the excluded members of society, such as individuals experiencing homelessness; this is carried out through information and education concerning these health issues (Information Principle of the Public Health Leadership Society) (25). In addition, we must resort to relational bioethics, specifically, to the concept that solidarity and care are moral practices (26). For Jennings, both solidarity and care imply the recognition of others, reaffirming the moral position of marginalized persons as members of society by recognizing their dignity and providing them health and social services according to their needs (26).

This would be reinforced by the ethics of care (27) within ethical caring, which arises in opposition to the lack of natural caring. Care

is associated with people's emotional relationships, which are distributed in concentric circles. At the most intimate level would be found the primary support networks in which care is established by affection. This is followed by the level of the close stranger or informal support and, last, by that of the remote stranger or help from society. People experiencing homelessness have lost their inner circle of care, while their informal supports are ambivalent and unstable due to their transitory situation.

In these cases, the ethical commitment to care extends not only to the State, but also to institutions and citizens, for the common good and solidarity, reflected in legislation as a guarantee of human rights.

3.2 Solidarity in public health policies

Currently, the term solidarity is commonly used to refer to the desire to promote the interests of others, even at personal cost (28). Solidarity understood in this way implies reciprocity, just as there are rights toward others, some obligations are also enforceable. From this perspective, investing in cancer prevention campaigns would lead to involvement in healthy lifestyles or, if not, exclusion from these programs. However, in public health, solidarity action cannot depend exclusively on reciprocity (29) because decisions about health are not isolated from the social context (30). This is the case in the prevention of cancer among people experiencing homelessness, whose choice of healthy lifestyles can be clearly limited by the social determinants they present and their life histories.

In this sense, solidarity implies the recognition of the moral identity of vulnerable individuals, reaffirming their position of equality in society. Solidarity and care implicitly recognize the other as a subject and help society provide resources and services to improve their health (26). In relation to medical care, Carol Goult identifies the structural injustices that still exist, even in solidarity health systems such as those in Europe (31). This recognition connects solidarity with justice (*solidarity of networks*), giving it a practical sense while positioning it as the need to fight to reduce social inequalities in health (31). This solidarity dispenses with taking measures that support those who have limited access to health services.

Their lack of economic resources deprives people of the possibility of achieving the capacities that are considered valuable, such as good health (32), which N. Daniels deems unacceptable and unfair (33). Poverty, homelessness, and discrimination based on race, gender, etc., are not isolated categories. Their intersectionality generates complex social inequalities. This is what happens to individuals experiencing homelessness, among whom poverty, mental health problems, damage related to substance use, racism, violence and cognitive disabilities intersect (34). This situation makes their access to health services even more difficult, which in the case of cancer implies an increase in morbidity and mortality, thus feeding back into their inequities.

Access to health services is key to reducing health inequalities (18). In the case of people experiencing homelessness, their higher mortality from cancer is not only the result of individual behaviors but is also related to their difficulty accessing cancer prevention programs (16). In this way, the responsibility for their disease is not only on these individuals but also on the functioning of the health systems.

In addition to interventions in social conditions, to advance equity, changes are needed in the health system that recognize and promote access to services for people with social vulnerability. This is the goal of the *CANCERLESS* project: supporting individuals experiencing homelessness so that they can have the same cancer prevention and screening opportunities as any other member of society.

However, on many occasions, the lack of public support limits the implementation of reforms aiming to reduce health disparities (35). At the base of this is what has been called the *status quo* bias, a position of aversion to change motivated by the benefits that individuals receive from the system without worrying about the damage they cause (36), justified in a liberal system, which considers social inequalities the product of the choice to lead an unhealthy way of life (37).

3.3 Relational autonomy for participation in cancer screening

It has often been suggested that in cancer screening programs, there is institutional pressure to increase the participation of individuals, with autonomy in decisions being underprioritized (38).

Autonomy has a strongly individualistic character, underscoring the decisions of people regardless of their circumstances, that is, their ability to exclusively make a choice without coercion or to make an informed decision. This notion has recently been questioned by bioethics following the impacts of the COVID-19 pandemic (39). Both concepts lack the relational sense that autonomy should have, considering the influence of social determinants on decision-making (40). Relational autonomy, although not yet well conceptualized, implies the recognition of the historical, social and cultural context of people making such a choice (41).

In this project, we suggest that decision-making on whether to participate in cancer screening programs should take into account the conditions in which people experiencing homelessness live. This implies respect for their values via a relational vision of autonomy that aims to involve participants in discussions about what best suits their personal preferences (42).

3.4 Social recognition and the virtue of just generosity

We cannot address the problem of social exclusion and foster the early detection of cancer among people experiencing homelessness, without rethinking ethics according to the verification of the fragility and exclusion of this population, specifically, by establishing that we are all interdependent in some stage of our lives.

MacIntyre argues that the virtue of just generosity is essential for knowing how to treat people who require support; it assumes that oneself has been, can be and will be a subject in need of care from others (43). Acting with just generosity requires that one operates via the attentive and affectionate consideration of the other. When this does not happen, it is always an indication of a moral defect, of an inability to act as duty requires.

Therefore, just generosity is not about helping people experiencing homelessness but concerns the recognition of their citizenship, ensuring that the State must be fair in the distribution of tasks to achieve this, thereby preventing the social abandonment suffered by individuals experiencing homelessness.

4 Conclusion

The ethical foundations of the *CANCERLESS* project are rooted in social justice and in equity in access to cancer screening programs for individuals experiencing homelessness.

The ethics of public health, originating in utilitarianism, are insufficient for serving the interests of the most disadvantaged groups in any population.

It is necessary to resort to a relational bioethics that includes solidarity and that recognizes the moral identity of socially excluded persons, reaffirming their position of equality in society.

The recognition that structural injustices still exist in health systems links solidarity with justice and positions it alongside the need to fight to reduce health inequalities.

Relational autonomy provides a broader conception of decision-making by considering the living conditions of people experiencing homelessness. Therefore, it is a more appropriate concept with regards to decision-making on participation in cancer screening programs. However, the State must generate possibilities in the distribution of such tasks to prevent their abandonment and to reduce the impact of this disease among the population experiencing homeless.

The recognition of citizenship and the virtue of just generosity can facilitate the equitable treatment of the population experiencing homeless, generating health systems focused on people that address their vulnerabilities.

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

MC-V: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. RG-T: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. BB-S: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. JB-C: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. AG-S: Validation, Visualization, Writing – original draft, Writing – review & editing. MA-P: Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. IG: Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Reexamining the use of race in medical algorithms: the maternal health calculator debate

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The concept of race is prevalent in medical, nursing, and public health literature. Clinicians often incorporate race into diagnostics, prognostic tools, and treatment guidelines. An example is the recently heavily debated use of race and ethnicity in the Vaginal Birth After Cesarean (VBAC) calculator. In this case, the critics argued that the use of race in this calculator implied that race confers immutable characteristics that affect the ability of women to give birth vaginally after a c-section. This debate is co-occurring as research continues to highlight the racial disparities in health outcomes, such as high maternal mortality among Black women compared to other racial groups in the United States. As the healthcare system contemplates the necessity of utilizing race—a social and political construct, to monitor health outcomes, it has sparked more questions about incorporating race into clinical algorithms, including pulmonary tests, kidney function tests, pharmacotherapies, and genetic testing. This paper critically examines the argument against the race-based Vaginal Birth After Cesarean (VBAC) calculator, shedding light on its implications. Moreover, it delves into the detrimental effects of normalizing race as a biological variable, which hinders progress in improving health outcomes and equity.

KEYWORDS

health disparities, VBAC, race, clinical algorithms, equity, maternal mortality

1 Introduction

The debate on racial categorization in healthcare persists, challenging the long-standing integration of race in medical diagnostics and treatment against the backdrop of social scientists' consensus that race holds no substantial biological foundation (1, 2). This divergence is starkly illustrated by the persisting health disparities across racial lines, with the alarming maternal mortality rates among African-American women—3.55 times higher than those of their White counterparts between 2016 and 2017—standing as a testament to systemic inequities (3). One emblematic medical algorithm case is the race-based Vaginal Birth After Cesarean (VBAC) calculator. This tool's potential biases may exacerbate health disparities by using race as a determinant in clinical decision-making, thereby reducing the likelihood that Black and Hispanic women will be recommended for Trial of Labor After Cesarean (TOLAC).

Medical algorithms are tools used to systematically approach clinical problems or treatment paths to reduce errors and improve healthcare (4). The VBAC calculator assesses the likelihood of a successful trial of labor after cesarean (TOLAC), factoring in BMI, maternal age, and previous cesarean details (5). However, its inclusion of race/ethnicity, despite aiming to refine predictions, risks reinforcing racial biases by suggesting unsubstantiated biological differences and, thus, potentially different care standards. However, its inclusion of race/ethnicity, despite aiming to refine predictions, risks reinforcing racial biases by suggesting

unsubstantiated biological differences. Associating race—a social construct—with innate biological capabilities, such as the ability to give birth vaginally after a cesarean section, not only perpetuates stereotypes but also potentially guides clinicians toward different standards of care based on race. This raises significant concerns about fairness and equity in medical practices. The debate surrounding the VBAC calculator highlights the challenge of leveraging technology to improve healthcare outcomes without perpetuating societal biases (6–8). Medical algorithms must be transparent, unbiased, and inclusive, minimizing past biases and prioritizing individual clinical characteristics to improve healthcare delivery (9).

This paper examines the push to phase out the race-based VBAC calculator, positioning this initiative within the broader discourse surrounding the intersections of technology, artificial intelligence, social justice, and the pursuit of equity in healthcare. It confronts the detrimental implications of conflating race with biological differences—a legacy rooted in the era of slavery and the historic exploitation of Black, Indigenous, and People of Color (BIPOC) in the development of science and medicine. Persisting in this practice not only deepens discriminatory patterns but also hinders progress in removing racism as a determinant of health outcomes (Jones, 2021). This exploration traces the origins of racial classification in American science and medicine, evaluates the debates surrounding the VBAC calculator, and critically examines the scientific and ethical underpinnings of race-based medical algorithms. This analysis advocates redefining race as a socio-political, rather than biological, category to foster a more equitable and just healthcare system.

2 History of racial categories in science and medicine

The idea of racialization (classifying people by race) appeared in English in the 1500s (10). In the 1700s, Carl Linnaeus, a taxonomist famous for categorizing plants and animals, proposed four classifications of humans based on skin color: Europaeus (white skin), Americanus (reddish), Asiaticus (tawny/tan), and Africanus (blackish) (11). Later, Blumenbach, a student of Linnaeus, divided humans into five groups based on geography and physical characteristics. In his classification, Caucasians were light-skinned people from Europe, and people living near Asia and Africa proximal to Europe were (Mongolians), Ethiopians (dark-skinned Africans), Americans (New World natives), and Malays (Polynesians) (12). Both Linnaeus and Blumenbach assumed a scientific stance, but their bias toward the social superiority of their European ancestry was evident in their writings. This presumption evolved into an established hierarchical order in which Europeans were at the top and Africans at the bottom, a moral justification for slavery, colonization, genocide, and discriminatory laws such as Jim Crow laws (13).

The concept of race was both politically and scientifically pivotal as the demographic composition of the United States evolved with the arrival of more immigrants. The first naturalization law in the United States, passed in 1790, restricted citizenship to "free white persons," thus institutionalizing racial categorization (14). Throughout history, the U.S. Census has played a significant role in shaping and reflecting categories of race. Initially, census enumerators identified individuals' race based on their perception, using categories such as "free White persons, enslaved people, or all other free persons" (15). As immigration and societal views on race changed, so did the categories, expanding to include mixed-race identifications such as mulatto, quadroon, and octoroon, and later detailed listings for Asian and Hispanic groups (15).

A significant change occurred in 1970 when the Census transitioned from enumerator identification to self-identification. This shift marked a crucial change in the control over racial identity as it allowed individuals to define their own racial identity. As a result of this methodological change, there were significant increases in the counts of some groups, particularly the American indigenous population (16). This shift emphasizes the fluidity of racial categories and highlights how they are influenced by social and political constructs rather than immutable biological differences.

The evolution of race statistics illuminates the utilization of physical appearances for categorizing individuals and as tools for political and social control. This reflects the broader dynamics of power and colonization that have shaped racial identities in America. The term "Indian" was initially used to "other" and marginalize the diverse Indigenous populations of America, further facilitating their exclusion from the nation-building process (15). As Irish immigrants and later other groups gradually assimilated into American culture, their racial categorization shifted, affording them the political and economic privileges reserved for Whites (17). This manipulation of racial categories to "other" various groups demonstrates how race was wielded as a tool for political and social control, showcasing the complex interplay of race, power, and identity in American history.

In science, the concept of race has been intertwined with ideologies of white supremacy, fueling movements that have led to the discrimination, elimination, and mistreatment of people of color under the guise of scientific advancement (18–20). Politicians such as Theodore Roosevelt and Winston Churchill supported the hypothesis of societal improvement using eugenics, a form of racial science focused on selective breeding and controlling human reproduction to achieve desired genetic traits (21). This highlights how genetics, as a scientific discipline, is an example of an area founded on racial ideology.

The renowned geneticist Francis Galton played a pivotal role in this history when he conceptualized eugenics in 1883. He described a range of physical, mental, and moral traits across races, arguing for the selective propagation of traits associated with the White race to improve societal health (22). This ideology was not isolated to genetics but extended into other scientific areas such as statistics. Figures like R.A. Fisher and Karl Pearson, credited with developing modern statistical methods, were deeply entrenched in the eugenics movement in England (23). They supported policies like sterilization of those with mental disabilities and race-based immigration controls.

In 1923, Henry F. Osborn, the then-president of the American Museum of Natural History in New York, publicly called upon the government to recognize the biological racial differences and preserve the virtue of the White race. Eugenics scientists received financial support from private donors, associations such as the American Breeders Association, and the government. The Eugenic Records Office in the Carnegie Institution was established by a prominent evolution scientist, Charles Davenport, who recruited Harry H. Laughlin as the superintendent of the ERO. The main goal of the ERO was to gather data supporting the eugenic movement and educate the public on the importance and implications of eugenic research. Laughlin was appointed as a congressional expert eugenics

agent by the US Congress Committee on Immigration. In 1922, he published a book on eugenic sterilization in the United States, arguing against integrating races and sterilizing individuals with mental disability (18). This contributed to state laws legalizing the sterilization of persons living with cognitive disabilities and later led to the mass forced sterilization of Indigenous and Black people in the United States (24).

These historical instances are a stark reminder of the enduring impact of race-based ideologies on shaping scientific thought and practice. By the early 20th century, the eugenics movement had grown into a significant scientific movement in the early 20th century, with American and European scientists embracing racial ideology as a science. They conducted experiments to propagate these false narratives and taught these concepts in universities, conferences, and even publicly (21). The legacy of these actions continues to influence the scientific landscape today, highlighting the crucial need for ongoing scrutiny and reform in how racial concepts are integrated into scientific research and discourse.

The embedding of eugenics in science created a significant challenge for cultural anthropologists and intellectuals, including W.E.B. De Bois and Franz Boas, who sought to counter the illogical racist theories. Franz Boas, widely regarded as a founder of modern cultural anthropology, employed scientific reasoning to refute earlier claims that Black people have smaller brains. For instance, he conducted a meticulous study measuring human skulls to provide evidence contradicting these assertions (25). On the other hand, Du Bois approached the issue of race from a social and historical perspective, viewing it as a mechanism used to group individuals and actively perpetuate economic and political oppression.

Eugenic policies were formally purged after World War II after the Nazi eugenics atrocities and the United Nations declared that race is a social construct (26). Though the United States denounced racial science officially, the ideology was already embedded into the power structures, particularly in science and immigration laws that favored White persons' immigration and discouraged interracial marriages. American publications of the American Eugenics Association and American Genetics Association fueled most Nazi eugenics atrocities (21). It is, therefore, no surprise that the idea of race science continues in scientific discourse and medical application of emerging genetic technologies that attempt to assign social and medical outcomes to immutable racial differences.

3 Evolution of racial categories in modern medicine

The early 2000s saw a revolutionary development in human genome sequencing, enabling a comprehensive insight into an individual's genetic makeup, encompassing variations, mutations, and potential disease markers within their DNA. Despite the scientific evidence, genetic similarities often surpass differences among individuals from different racial groups (11, 27). Furthermore, the persistence of racial biological essentialism, characterized by the belief in race as a biological and genetic entity, continues to have significant social and political ramifications. This enduring notion, despite its scientific debunking, underscores how deeply ingrained and complex the issue of race remains in our society, influencing both societal interactions and governmental choices.

An illustrative example is the US Census and Vital Statistics, which collects racial data corresponding to ancestry or geographical region- an adaptation of Blumenbach's racial classification (see Figure 1). Since the 1980s, the US has also measured "Hispanic" as an ethnic group of people who speak Spanish and originate from Mexico, Puerto Rico, Cuba, Central and South America, and other Spanish-speaking countries except for Portugal and Brazil (13). Definitions of race and ethnicity have evolved to reflect cultural and social norms (13). Ethnicity, which may overlap with race, is a subjective label for people who share cultural, language, or physical attributes (11). Similar to race, ethnicity is a social construct with complex and fluid dimensions that are difficult to measure scientifically. So, in the case of VBAC, how did race (being "Black") and ethnicity (being "Hispanic') become negative variables in a clinical tool?

4 Case study: the VBAC calculator and maternal health

Cesarean delivery, an abdominal surgery for childbirth, carries various risks that can affect both mother and child, potentially leading to increased rates of mortality and morbidity (28). Over the past few decades, cesarean rates in the United States have surged, reaching 32% by 2009 (28). Traditionally, women who had undergone a cesarean were often expected to repeat the procedure for future births. This changed in the mid-1980s, as evidence emerged suggesting that Vaginal Birth After Cesarean (VBAC) could be a safe alternative for certain patients, offering reduced risks associated with repeat cesareans, thus prompting a shift toward encouraging Trial of Labor After Cesarean (TOLAC) (29).

In 2007, the Maternal-Fetal Medicine Units (MFMU), supported by the National Institutes of Health, created a VBAC calculator to aid healthcare providers in evaluating the viability of TOLAC for individual patients. This tool, which received endorsement from the American College of Obstetrics and Gynecologists, considers several factors, including body mass index (BMI), patient age, cesarean history, and race/ethnicity, to predict the success of VBAC (6, 30).

The VBAC calculator decreased the likelihood of VBAC success for women identified as African American/Black and Hispanic (Table 1). Using the assumption that Black and Hispanic women have less successful VBAC, the calculator subtracted from the score, which gave these women less chance of TOLAC. Consequently, women with the same age, BMI, and history of cesarean had different scores based on their identified race/ethnicity. Vyas et al. (6) challenged the VBAC's use of these race-based correction factors. They argued that if scores influenced clinicians' decisions, the calculator probably contributed to maternal disparities (6). Vyas (31) also noted that many other factors, such as marital status and insurance, were identified in creating and validating the tool that could have been incorporated into the predictive tool used in the United States, Israel, Italy, United Kingdom, Netherlands, and New Zealand (32). In fact, the version of the tool used in most countries does not include race correction. Therefore, embedding race/ethnicity corrections in the US-based VBAC creates inequitable treatment by race and further propagates the notion that racial disparities are immutable.

The Maternal-Fetal Medicine Units Network (MFMU), which developed the race-adjusted VBAC calculator, intended to aid in clinical decision-making rather than to perpetuate discrimination

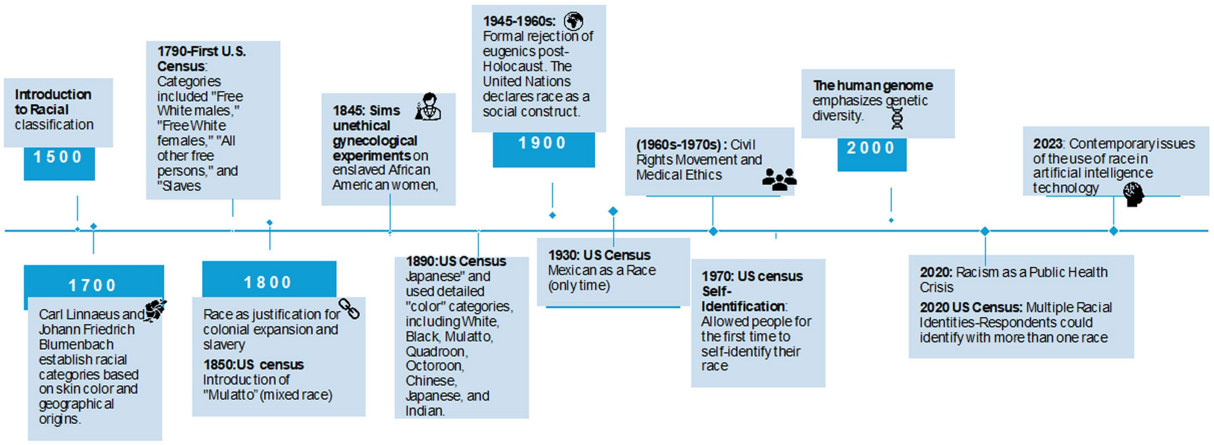

FIGURE 1
Chronology of racial categorization and its impact in medicine and society. The timeline represents a selection of key events that have influenced the concept and application of racial categorization in medicine and its broader societal implications. It is not exhaustive but highlights pivotal moments shaping current perspectives on race in medical practices and technologies. The historical "color" categories used in the U.S. Census reflect the sociopolitical constructs of race during specific periods and may be considered outdated and insensitive today. "White" referred to individuals of European descent; "Black" to those of African descent; "Mulatto" indicated mixed African and European ancestry; "Quadroon" and "Octoroon" described individuals with one-quarter or one-eighth African ancestry, respectively. "Chinese," "Japanese," and "Indian" referred to individuals from those respective ethnic or national origin.

TABLE 1 Comparison of VBAC success rate calculations with and without race/ethnicity.

Input criteria	With race and ethnicity	Without race/ethnicity
Patient's age	✓	✓
BMI	✓	✓
Previous cesarean	✓	✓
Race/ethnicity	✓	X
Sample predicted VBAC score	48.2% success rate (95% confidence interval 44.5, 51.9%)	64.5% success rate (95% confidence interval 62.1, 66.9%)

The sample VBAC success rates above were calculated for a hypothetical patient profile with the following characteristics: Maternal Age of 31 years, BMI of 22, no prior vaginal birth, and a previous cesarean section due to arrest of dilation. The rate for the African American/Hispanic column incorporates race/ethnicity as a risk factor, as per the original VBAC algorithm, whereas the rate for the Non-African American/Hispanic column excludes this variable, reflecting the updated calculation approach. The confidence intervals provide a statistical estimate of the range in which the actual success rate may fall.

against Black and Hispanic women. Nonetheless, the calculator's race adjustments resulted in lower estimated VBAC success probabilities—by 5–15 percentage points—for Black and Hispanic patients compared to White patients with similar clinical profiles, based on analyses from a large cohort study with an evidence level II (30). Furthermore, their model suggested that patients with scores below 60 percent might reduce morbidity by opting for a repeat cesarean over attempting a VBAC (33). However, this calculator's utilization of race as a biological variable is emblematic of a broader trend within epidemiological research. Historically, race and ethnicity have been employed as imprecise surrogates for complex social and health factors, thereby perpetuating a systemic issue where the scientific application of race may conceal actual social determinants of health. For example, factors such as differential access to healthcare, environmental exposures, and socio-economic disparities are critical but are often masked by the simplistic categorization by race (34). This reductionist approach can lead to misdiagnoses and inequitable health outcomes, as it overlooks the multifaceted nature of health determinants, such as the impact of living in high-pollution areas or the chronic stress associated with racial and economic marginalization (35).

The subsequent racial discrimination controversy surrounding the VBAC calculator prompted a reevaluation, leading Grobman and colleagues to revise the tool, replacing race/ethnicity with medical history components such as hypertension (5). This incident has sparked broader discourse on the critical need to reassess the role of race in clinical algorithms and to acknowledge the potential biases that arise from its misuse (31, 36).

5 Legacy of the VBAC calculator

The history of science and technology, such as genetics, shows how ideology influences science. In general, it is assumed that there is a separate scientific meaning of race in science that is not contaminated by the sociopolitical meaning of race. Roberts (1) argues that the biologization of race (use of race as an inherent biological fact) is acceptable today because racism is normalized, making it invisible. In medicine, there are existing race-based guidelines taught to clinicians currently in use. In a recent article, Amutah et al. (37) presented a case of a patient of mixed parental ancestry being considered for a kidney transplant. Given the existing race correction adjustment for Black patients in the glomerular filtration rate (GFR), the patient has differential access to the transplant list depending on which race he is considered to be.

In this kidney function test scenario, the racial differences come from a racist presumption that Black patients have greater muscle mass than other races; therefore, the GFR needs to be adjusted (38).

These beliefs of Black individuals having denser muscle and thicker skin have been used to justify harmful medical practices. For example, in the 1950s, Black patients were dosed with higher X-ray radiations based on unsubstantiated beliefs grounded on racism (39). In this case, White individuals are assumed to be "normal," and Black individuals or people of color need more X-rays to penetrate their skin. Similarly, in pain medicine, there has been an assumption that Black people have thick skin and feel less pain. This has led to the mistreatment of Black patients in medicine, including performing surgery without anesthesia.

In obstetrics, studies conducted in the 1920s relied on the racialized anatomy concepts published and propagated notions that White women had a standard pelvis ideal for childbirth. In contrast, Black and Indigenous women were assumed to be anatomically deficient (19). These assumptions of faulty anatomy led to high rates of interventions such as cesareans among women of color to compensate for their abnormalities (40). Additionally, these notions of the inferiority of Black and Indigenous women were used to justify forced sterilization (41). Despite the obvious racist antecedents of the inferiority of women of color pelvic anatomy, researchers continue to cite racial and ethnic variation in pelvic as a factor contributing to adverse childbirth outcomes (6).

6 Policy implications and future directions

A recent draft guideline from the National Institutes of Health and Care Excellence (NICE) suggests that labor induction should be considered at 39 weeks for women of Black, Asian, or minoritized background, even if the pregnancy is uncomplicated (42). The guideline recommends labor induction for White women at 41 weeks of gestation. Clinicians who argued against the recommendation noted that race has been used as a proxy for social and medical factors (43). Again, in this case, there is an implicit presumption that race confers immutable characteristics.

It is not scientifically accurate to use broad racial categories like Asian or African in clinical decisions. For instance, people categorized as Asians comprise a diverse group of individuals, including Chinese, Japanese, Indians, Filipinos, Thais, and others. This is a large geographical region with various social and cultural factors that impact health. Additionally, DNA sequencing has revealed significant variability within African populations. Thus, grouping Africans into one category does not make any biological sense. Race and ethnicity are fluid social constructs and unreliable indicators of ancestry or genetics.

The examples of VBAC and NICE guidelines' scrutiny of the use of race illustrate a critical need to reexamine the institutionalization of racism in medicine. A recent systematic analysis of UpToDate articles showed that for articles that mention race, biologization of race occurred in 93.3% of the articles, and there were discussions of inherent racial differences without context (44). Furthermore, 32.7% of the articles racialized biomedical research and clinical practice. This included references to racialized patterns of behavior and cultural practices. There was also insufficient data on Black populations, limiting the study to a specific racial group and race-based clinical practice guidelines. The widespread use of these articles in clinical decision-making among clinicians and medical and nursing students raises the question of whether the normalization of race can be systematically dismantled.

In a recent article in Pediatrics, Wright et al. (45) argued that evidence from the human genome project, stress, and adaptation studies provide enough evidence to dismantle race-based medicine. Similarly, other researchers and clinicians have concluded that race is inaccurate in understanding human diversity and clinical race-based predictions (31). However, as Vyas et al. (31) explained, a lack of evidence of genetic races has not stopped the belief from manifesting insidiously in clinical practice. This belief is also true in the American general public. For example, a recent poll showed that differences in the socioeconomic status of White and Black people were due to genetics (27). These essentialist theories, especially among White individuals, reduce the support for policies that attempt to dismantle systemic racism- a social determinant of health.

Nonetheless, only a few systematic solutions are proposed apart from the slow progress of undoing race corrections one at a time. Kane et al. (36) proposed that clinicians and researchers use structurally just algorithms prioritizing social drivers of inequities such as insurance status, education, and economics. This alternative approach emphasizes structural justice by analyzing the root causes and working collaboratively with advocates and communities to address societal-level circumstances contributing to disparities such as those noted in maternal and child mortality. In addition, it may be prudent to return to the drawing board and decenter diagnostics, prognostic tools, and treatment guidelines from one racial group and instead create an inclusive approach to biomedical research and healthcare.

7 Conclusion

The history of science and technology shows that social ideologies influence science. Therefore, contrary to the prevalent use of race as a biological variable, evidence shows that genetic and biological races do not exist. The acceptability of race in medicine is particularly troubling since it has caused iatrogenic harm and possibly exacerbates health disparities. In the case of VBAC, creating medical algorithms that discriminate against Black and Hispanic women based on race and ethnicity (social constructs) detracts from efforts to improve maternal mortality. Further, using race causes harm by miscategorizing people based on fixed ideas of race, reinforces biological essentialism, and prevents support for reforms to eliminate racism as a social determinant of health.

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

RK: Conceptualization, Formal analysis, Methodology, Visualization, Writing – original draft, Writing – review & editing.

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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.

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Impact of urban and rural residents medical insurance on self-rated health of residents in China: a panel study from the China family panel studies national baseline survey

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Objective: This study aimed to investigate the health performance of the Urban and Rural Residents Medical Insurance (URRMI) scheme in China and to make practical recommendations and scientific references for its full implementation in China.

Methods: This is a panel study that uses data from the China Family Panel Studies from 2018 to 2020, which is separated into treated and control groups each year, utilizing the key approach of propensity score matching and difference-in-difference (PSM-DID). Using 1-to-1 k-nearest neighbor matching, we proportionate the baseline data. Using difference-in-difference model, we examine the mean treatment impact of the outcome variables. Using a 500-time random sample regression model, we validate the robustness of the model estimation.

Results: The result was credible after matching, minimizing discrepancies. Good overall performance of self-rated health with an average Hukou status of, respectively, 0.8 and 0.4 in the treated and control group, primarily in rural and urban regions separately. The participation of URRMI significantly impacted self-rated health of residents, with a 0.456-unit improvement probabilities observed ($p < 0.1$). Additionally, the individuals are categorized into urban and rural, and those with urban hukou had a 0.311 expansion in the probability of having better health status compared to rural hukou ($p < 0.05$). Other factors, such as age, highest education, annual income, medical expenditure, hospital scale, clinic satisfaction, and napping, also impacted self-rated health. Moreover, elder individuals, higher education levels, and higher medical expenditure having a higher probability of improvement. The study utilized a placebo test to verify the robustness of the URRMI regression. The estimated coefficients showed that basic medical insurance did not significantly improve the health of insured residents under the URRMI scheme.

Conclusion: The study demonstrates the crucial role of PSM-DID in determining the influence of URRMI on self-rated health status. It indicates that purchasing in URRMI has a favorable influence on the health of residents, advancing enhanced self-rated health effectiveness. It does, however, reveal geographical disparities in health, with urban dwellers faring far better than those who live in the suburb. Study suggests expanding URRMI coverage, narrowing urban–rural

divide, increasing insurance subsidies, reforming laws, and developing effective advertising strategies.

KEYWORDS

urban and rural resident medical insurance, health performance, resident, selfrated health, propensity score matching and difference-in-difference regression

1 Introduction

The World Health Organization (WHO) implemented Universal Health Coverage (UHC) in 2015, which ensures that populaces have admittance to the health care they require exclusive of incurring financial difficulty. UHC is one of the sustainable development goals (SDGs) approved by the world's states in 2015. The WHO also proposed the UHC2030 plan, which is a global campaign to establish strong health systems to achieve UHC. The approach to Health Systems Strengthening (HSS) is crucial for achieving UHC. The healthcare system is generally understood as all authorized public and private organizations, institutions, and resources that are responsible for improving, maintaining, and restoring health. HSS entails investment in participation in an incorporated and systemic approach and altering the architecture that governs how various components of the health system operate and interrelate to address primary health needs through people-centered amalgamated services (1). As a result, HSS is the primary means of achieving UHC.

The Chinese government has always prioritized the health of its citizens, demonstrating a commitment to improving the accessibility to healthcare and affordability of medical costs for all in its programs. A paramount goal of the government is to achieve UHC of the basic social medical insurance system and to relieve the financial burden associated with medical treatment.

The basic social medical insurance system in China comprises the NRCMS in rural areas and the UEBMI and URBMI in urban areas. The basic social medical insurance system in China comprises the New Rural Cooperative Medical Scheme (NRCMS) in rural areas, and the Urban Employee Basic Medical Insurance (UEBMI) and Urban Resident Basic Medical Insurance (URBMI) in urban areas. In order to better solve the problem of medical resource imbalance caused by the urban–rural dual structure in China, the Central Committee of the Communist Party of China proposed the integration of the Urban and Rural Residents Medical Insurance System (URRMI) in November 2013, which was officially implemented in 2016.

With the swift economic expansion and appropriate improvement of medical and health facilities in China, the social security system has gradually begun to detach from construction, imposing a severe financial burden on the masses seeking medical treatment. By 2000, the proportion of personal health expenditure borne by Chinese residents had peaked, surpassing 60.6% of the overall health expenditure in China (2), and illness-related poverty had become a leading driver of poverty. Since the late 1990s, medical and health system reform has focused on constructing and improving a basic social medical insurance system. In 2013, China achieved complete coverage of the basic medical security system. By the end of 2021, the number of Chinese dwellers on national basic medical insurance was 1362.97 million, with a participation percentage of more than 95% (3). The proportion of China's total health expenditure directly born by individuals and households has decreased annually, from 60% in 2001 to 27.7% in 2021 (4), with improvements in population coverage and security. Additionally, the problematic of "kan bing nan, kan bing gui," which means the difficulty and high cost of getting medical treatment, has been significantly alleviated.

The UEBMI system was first designed to advance the coverage of urban medical insurance in China. It is a social security scheme designed to compensate urban employees and pensioners but not their families for economic losses caused by disease risk, introduced in two medium-sized cities (Zhenjiang and Jiujiang) in 1994 and expanded countrywide in 1998 (5). It went into effect in 1998 to cover urban employees, while the URBMI was implemented in 2007 to cover urban inhabitants (6). The UEBMI benefit packages are envisioned to cover not just inpatient medical care but also outpatient facilities, such as medical treatment for serious and chronic diseases (7). By the end of 2021, the number of employees enrolled in medical insurance was 354.31 million, a 9.76 million rise and a 2.8% increase over the previous year (3).

The NRCMS is a government-led voluntary insurance system established in 2003 to increase access to health insurance for rural citizens (8). Unlike obligatory insurance, the NRCMS is run and administered by the county. The central government connected the delivery of its subsidies to the extent of coverage in each country, providing local governments with a strong incentive to expand coverage. Enrollment in the NRCMS is typically centered on households rather than individuals, which is one of the most effective strategies for quick coverage expansion (9). The URBMI was launched with significant government subsidies and is designed similarly to the NRCMS. Its primary beneficiaries are children, the older adult, college students, and unemployed urban dwellers who are not covered by the UEBMI plan (10). After the implementation of the basic medical insurance system for urban employees and the new rural cooperative medical care system, it is a major initiative taken by the CPC Central Committee and the State Council to further address the healthcare security issues of the broad masses of people and continuously improve the healthcare security system. It mainly makes institutional arrangements for non-employed urban residents' medical insurance. The introduction of this system is of great significance in the process of China's social insurance system reform, indicating the direction of China's social insurance system reform.

Abbreviations: WHO, World Health Organization; UHC, Universal Health Coverage; HSS, Health Systems Strengthening; NRCMS, New Rural Cooperative Medical Scheme; UEBMI, Urban Employee Basic Medical Insurance; URBMI, Urban Resident Basic Medical Insurance; URRMI, Urban and Rural Residents Medical Insurance System; CFPS, China Family Panel Studies; PSM-DID, propensity score matching and difference-in-difference; LTCI, long-term care insurance.

Following the incorporation of the URBMI into the NRCMS in 2016, China has founded a basic social medical insurance system, with the main entities being the UEBMI and the URRMI schemes. The merger of both schemes has improved the justice of the basic medical insurance system. By enrolling in a cohesive medical insurance system for urban and rural residents, residents can more fairly enjoy basic medical security rights and interests in accordance with the unified policy of insurance payment and treatment (11). The insurance benefits became more balanced following the merger and integration. Anyone with a recognized permanent residence in the countryside can be treated the same as those in cities. This equitable approach makes medical insurance reimbursement more practical and enables the basic medical "service packages" that the general public can access to be upgraded to meet higher standards. The basic medical "service packages" include expanding medical insurance reimbursement and broadening the variety of drugs enclosed in medical insurance. The use of URRMI enables the implementation of integrated handling service management, making management more unified. It removes barriers such as urban–rural system separation, management segmentation, and resource dispersion, making the transfer and linkage of medical insurance relationships amid urban and rural inhabitants more convenient.

Due to differences in healthcare insurance systems across countries, such as the commercial healthcare insurance system in America, the national health service system in Britain, and the social medical insurance system in China, it can still be found that healthcare insurance can provide certain assistance to the health of policyholders. Shi et al. from America investigated the association among insurance rank and general survival in female breast cancer patients attending public hospitals and observed that uninsured individuals outlive insured patients (12). Using data from groups in U.S. states, Thornton et al. observed that participation in commercial health insurance could save more than 75,000 lives per year by improving population health outcomes while extending commercial health insurance coverage to all uninsured people in the United States (13). A study showed that after 1 year of enrollment, enlarging health insurance had momentous effects on self-reported, mental, and physical health, although there was no substantial effect on humanity during the observation period (14). Another study using nationwide illustrative statistics from the Demographic and Health Surveys of Ghana, Indonesia, and Rwanda found that expanding health insurance to involve income-sensitive extras or exemptions for people with low income, as well as low or no copayments, can surge the usage of affectionate health care (15).

Chinese researchers also did numerous empirical research on health performance, exclusively concentrating on basic medical insurance. Corresponding to a study that used figures from the China Health and Nutrition Survey, participation in the new agricultural cooperation reduced the pervasiveness of some diseases, increased the pervasiveness of injuries and various diseases, and significantly improved participants' self-rated health status (16). Shanshan et al., using this data analogously, observed that the new rural cooperative and urban housing insurance had no obvious positive effect on children's short-term health, but the new agricultural cooperative had a meaningful constructive effect on children's long-term health and significantly improved their health status (17). Based on data from the two phases of the China Health and Retirement Longitudinal Study, Lianjie et al. determined that integrating medical insurance for urban and rural inhabitants promotes the physical and mental health of older adult individuals in rural regions by increasing medical service consumption (18). Another study evaluated the temporary and continuing health evaluation indicators and found that the basic medical insurance of urban employees increased the actual medical costs of the insured population (19). However, it also had an optimistic effect on the health of the insured population or produced positive health performance (19). Meng et al. used statistics from the 2015 China Migrants Dynamic Survey to study the senior floating population and found that involvement in the health insurance system considerably improved the self-rated health of floating seniors (20).

Studies on the quantitative evaluation of medical insurance have some limitations. First, most studies rely on prevalence and mortality as evidence of the health effects of insurance, and there is less discussion about the causal link between self-rated health status and registration in insurance. Second, uncertainties persist concerning the effect of medical insurance on health, with some studies demonstrating no significant positive effect. Finally, most analyses have focused on the positive health promotion effect of health insurance, but the researches on URRMI insurance are limited. According to the health production theory of health economics, numerous factors influence health, including genetic inheritance, healthy behavior, dietary status, living environment, and medical services. The connection concerning health insurance and health is complicated because, under the idea of voluntary insurance, insurance status is frequently the product of human choice and an endogenous variable. Therefore, identifying the causative connection concerning health insurance and health status voguish studies is challenging.

This study used data from the large-scale China Family Panel Studies (CFPS) from 2018 to 2020 and propensity score matching and difference-in-difference regression (PSM-DID) to evaluate the impact of URRMI on the health utilization of urban and rural residents. We aimed to specify some recommendations on the policy of completely adopting the URRMI system in China, in addition to their practicability.

2 Materials and methods

2.1 Data source

This study used data from the CFPS database, which was conducted in 2010 by the Chinese Peking University's China Social Survey Center. The survey was organized into three sections: individual, family, and community, and it used stratified multi-stage sampling to collect data from 25 provincial administrative units. The poll covered various topics, including economic activity, educational success, family dynamics, population movement, and health. The information is reliable and true. Follow-up data of the CFPS database from 2018 and 2020 were used as experimental samples in this investigation. A final sample of 7,364 individuals was included in the regression after selection, and then separated into URRMI group (treated group) and non-URRMI group (control group) (Figure 1), including the sample data from the point where the two data periods intersected.

2.2 Model method

This study engrossed 1:1 k-nearest neighbor matching to determine how URRMI affected the self-rated health of Chinese individuals. A

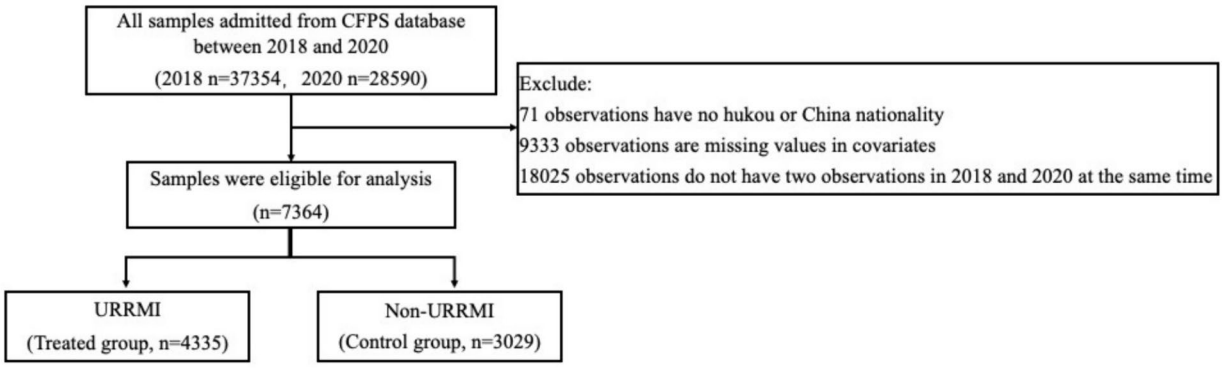

FIGURE 1
Study design and flow chart of the observations collection and the classification of observations with and without urban and rural residents medical insurance (URRMI) for propensity score matching. The n stands for the observations from 2018 and 2020, respectively. CFPS, China Family Panel Studies.

comparative experiment was designed to identify the unique effects of URRMI on individual self-assessment by comparing the self-rated health levels among the treated and control groups. This improved the hypothetical self-selection bias of the samples. When evaluating the health effects of URRMI on residents, it is crucial to account for selectivity bias based on observable variables like age, wealth, and family status produced by residents' voluntary choice of insurance. We created a suitable control group using the PSM to avoid selectivity bias. PSM is frequently used to calculate the effects of health and other policy initiatives since randomized controlled testing is impractical (21).

The DID model was used for further analysis after resolving the problem of individual self-selection. Therefore, selectivity bias due to subjective observation and time-varying traits can be successfully mitigated using the variance. The difference between the average change in self-rated fitness levels in the experimental group from 2018 to 2020 was also compared based on PSM. Finally, the average treatment effect for the treated group (ATT) was obtained after eliminating selectivity bias from observable and unobservable characteristics.

2.3 Variables

2.3.1 Explained variable

The explained variable of this study is self-rated health, which assesses the health level of the individual. It was coded as a two-valued dummy variable with a health status of unhealthy or average, defined as 0 = unhealthy, and a health status of relatively healthy, healthy, and very healthy, defined as 1 = healthy.

TABLE 1 Grouping information for the treated group and control group in 2018/2020.

	2018	2020
treated group	not purchase URRMI (included URBMI or NRCMS)	purchase URRMI (included URBMI, NRCMS, or URRMI)
control group	not purchase URRMI (included URBMI or NRCMS)	not purchase URRMI (included URBMI, NRCMS, or URRMI)

2.3.2 Explanatory variables

The explanatory variable was the URRMI scheme. Since 2016, the URRMI Scheme has gradually amalgamated the URBMI and the NRCMS. Due to the questionnaire design of CFPS database, individuals who participated in URBMI or NRCMS were considered part of the URRMI for the 2018 statistics. Therefore, individuals who participated in URBMI, NRCMS, or URRMI were included in the URRMI for the 2020 statistics. People who did not purchase URRMI in 2018 and those who purchased URRMI in 2020 were included in the treated group. The control group consisted of people who did not purchase URRMI in both 2018 and 2020, as reflected by the grouping dummy variable treated (Table 1). However, because this study's model is a double difference model, the product of the interactive term of the time and the treated group dummy variables (Time*Treated) was the primary explanatory variable to effectively quantify the impact of urban and rural basic medical insurance on the health of residents.

2.3.3 Covariates

In this study, we used the Grossman model (22) as a template and selected three levels of control variables based on relevant literature (23–25): (1) Individual characteristics: Hukou status, Age, Gender, Marital status, Highest education, Annual income; (2) Selection of medical and health services: Hospitalization, Medical expenditure, Hospital scale, Clinic satisfaction, Hospital quality; and (3) Health behavior: Smoking, Drinking, and Napping.

Regarding individual characteristics, Hukou status was coded as 1 = rural hukou and 0 = urban hukou. Age and Gender were not treated, based on the original. Marital status was defined by the presence or absence of a spouse. Individuals who were married or cohabiting were considered to have a spouse, while those who were unmarried, divorced, or widowed were defined as not having a spouse. The variable Highest education was treated similarly, retaining the individuals who had primary education, junior secondary education, or senior secondary education. Individuals who were not in school and illiterate/semi-literate were merged into the illiterate/semi-literate category. Additionally, junior college, undergraduate degree, master's degree, and doctorate were classified as higher education. The observed values of Annual income were log-transformed, basing on the actual income data. Regarding medical and health services, Hospitalization,

TABLE 2 Variable assignment of covariates.

Variables	Variable assignment
Individual characteristics	
Hukou status	1 = rural hukou 0 = urban hukou
Age	the actual age of the samples
Gender	1 = male 0 = female
Marital status	1 = married 0 = unmarried
Highest education	0 = illiterate/semi-literate category 1 = primary education 2 = junior secondary education 3 = senior secondary education 4 = higher education
Annual income	the actual annual income of samples with log-transformed
Selection of medical and health services	
Hospitalization	1 = yes 0 = no
Medical expenditure	actual medical expenditure of samples with log-transformed
Hospital scale	1 = general hospital 2 = specialist hospital 3 = community healthcare center/ township hospital 4 = community healthcare clinic/village clinic 5 = clinic
Clinic satisfaction	1 = excellent 2 = very good 3 = good 4 = poor 5 = disappointed
Hospital quality	1 = excellent 2 = very good 3 = good 4 = poor 5 = disappointed
Health behavior	
Smoke	1 = yes 0 = no
Drink	1 = yes 0 = no
Nap	1 = yes 0 = no

Hospital scale, Clinic satisfaction, and Hospital quality were not processed, while Medical expenditure was log-transformed. According to the questionnaire, Hospitalization is a binary question, whereas it also incorporates five categories of Hospital scale and three different attitudes toward Clinic satisfaction. Regarding health behaviors, Smoke status was defined as smoking, which included quitting smoking. Drink status and Nap were not treated. Following the questionnaire, we have defined Drink status as indicating whether an individual has consumed alcohol three times a week in the past month, and Nap as indicating whether an individual has taken the nap. The assignment and definition of each variable are outlined in Table 2.

2.3.4 Statistical analysis

This study started with PSM therapy. The projected likelihood of each sample participating in URRMI was calculated using treated as the explanatory variable, a logit model for regression was built, and a new set of observation samples were created by matching using the 1:1 nearest neighbor matching approach. The model settings are shown in Equation 1:

$$logit\left(Treated_i = 1\right) = \gamma_0 + \gamma_1 X_i + \varepsilon_{it} \quad (1)$$

where i represents the individual. *Treated* indicated participation in URRMI and was assigned a value of 1 if the individual participated and 0 otherwise. X_i is the matching variable, which included Hukou status, Gender, Marital status, Annual income, Hospitalization, Medical expenditure, Hospital scale, Clinic satisfaction, Hospital quality, Smoke, Drink, and Nap. ε_{it} expresses error terms and contains information other than the main variables of the model. The current study aims to identify, for each individual in the treated group who purchased URRMI, a matched control individual in the same year who did not secure URRMI, and to eliminate samples that were not magnificently matched. The matched statistics will then be composed year by year and a DID regression will be presented on the combination of general statistics.

The following equation of the DID model was estimated for continuous outcomes:

$$Health_{it} = \alpha_0 + \alpha_1 Treated_{it} + \alpha_2 Time_{it} + \alpha_3\left(Treated_{it} \times Time_{it}\right) + \beta_i \times X_{it} + \varepsilon_{it} \quad (2)$$

Where $Health_{it}$ stands for self-rated health of resident i at time t; the treated variable $Treated_{it}$ is a binary indicator; it epitomizes the group dummy variable. $Treated_{it} = 1$ indicates that an individual i belongs to the treated group, having participated in the URRMI. $Treated_{it} = 0$ epitomizes the control group, indicating that the individual i did not participate in the URRMI. $Time_{it}$ epitomizes the time dummy variable, with $Time_{it} = 0$ indicating the time t before the individual i participated in the URRMI (the year 2018) and $Time_{it} = 1$ indicating the time t after the individual i participated in the URRM (the year 2020). The variable $Treated_{it} \times Time_{it}$ signifies the collaboration among groups and time. X_{it} epitomizes a set of individual covariates of individual i at time t. ε_{it} expresses error terms and contains information other than the main variables of the model.

Stata 15.0 in Mac was used for statistical cleaning analysis. The two-sided statistical significance level was set to 0.05.

3 Results

3.1 Samples characteristics

After statistic cleaning and PSM matching, we analyzed a total of 2,021 treated group samples and 1,372 control group samples from

TABLE 3 Characteristics of the study sample in the CFPS 2018 and 2020.

Variables	2018				2020			
	Treated		Control		Treated		Control	
	(*n* = 2,021)		(*n* = 1,372)		(*n* = 2,314)		(*n* = 1,657)	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD
Health	0.78	0.42	0.81	0.39	0.75	0.43	0.80	0.40
Individual characteristics								
Hukou status	0.77	0.42	0.41	0.49	0.82	0.39	0.37	0.48
Age	41.25	12.83	38.33	11.35	40.23	12.62	38.60	11.42
Gender	0.56	0.50	0.55	0.50	0.57	0.50	0.56	0.50
Marital status	0.79	0.41	0.75	0.44	0.83	0.37	0.78	0.42
Highest education	1.36	1.20	2.53	1.30	1.20	1.07	2.47	1.29
Annual income	10.09	1.03	10.71	0.91	9.98	0.95	10.53	0.82
Selection of medical and health services								
Hospitalization	0.12	0.32	0.10	0.29	0.12	0.32	0.10	0.30
Medical expenditure	7.24	1.67	7.27	1.61	7.20	1.57	7.26	1.53
Hospital scale	2.95	1.63	2.32	1.62	3.09	1.62	2.29	1.61
Clinic satisfaction	3.72	0.77	3.74	0.74	3.56	0.82	3.58	0.80
Hospital quality	3.56	0.91	3.64	0.86	3.39	0.86	3.46	0.84
Health behavior								
Smoke	0.59	0.49	0.63	0.48	0.54	0.50	0.59	0.49
Drink	0.14	0.35	0.13	0.33	0.16	0.37	0.15	0.36
Nap	0.62	0.49	0.65	0.48	0.51	0.50	0.58	0.49

2018, as well as 2,314 treated group samples and 1,657 control group samples from 2020 (Table 3). The overall self-rated health of the samples was good, with each average health status above 0.78, 0.81, 0.75, and 0.80. Comparing the standard deviations of the four groups, it can be seen that the differences among the four groups are not significant (standard deviation [SD] 0.42, 0.39, 0.43, and 0.40, respectively). The average Hukou status of both two treated groups was approximately 0.8, concentrated in rural regions. In contrast, the average Hukou status of both two control groups was less than 0.5, with the majority of individuals living in urban regions. The average age of the 2018 treated group was 41.25 years (SD 12.83), while the average age of the 2018 control group was 38.33 years (SD11.35). Similarly, the average age of the 2020 treated group was 40.23 years (SD12.62), and the average age of the 2020 control group was 38.6 years (SD11.42). The gender distribution was relatively balanced within each group, and the differences between groups were not significant. The average Marital status of all four groups was above 0.5, indicating that the majority of participants were married. The average for the Highest education was around 1.3, indicating that the majority of individuals had completed compulsory education. Similarly, there were no significant differences in Annual income between the four groups. The average Hospitalization score for each group was 0.12, 0.1, 0.12, and 0.1, respectively (SD 0.32, 0.29, 0.32, and 0.3, respectively), indicating that the majority of participants had not been hospitalized due to illness. The average Hospital scale for each group was 2.95, 2.32, 3.09, and 2.29, respectively (SD 1.63, 1.62, 1.62, and 1.61, respectively), indicating that the majority of participants had been treated at specialist hospitals or community health centers. The average Clinic satisfaction was between general and satisfied. The average Hospital quality was between general and good. The average Smoke for each group was above 0.5, with the 2018 control group having a score of 0.63, indicating that smoking behavior was prevalent. The average of Drink for each group was around 0.1, indicating that drinking behavior was less common. The average Nap score for each group was around 0.6, with the 2020 experimental group being close to 0.5, indicating that the differences between individuals who habitually take naps and those who do were not significant, but there were slightly more individuals who did not habitually take naps.

3.2 Balancing proper test for PSM results

The aforementioned model of Equation 1 was used to determine PSM. The balancing characteristics of the observable covariates were assessed between the treated groups and control groups using k-nearest neighbor matching to reduce sampling bias (Table 4). Unmatched refers to the treated group and control group samples before matching, whereas matched refers to the treated group and control group samples with approximately constant distribution following propensity matching.

Age, Medical level, Smoke, and Nap were significantly different before and after matching. Rosenbaum argues that the matching result is reliable when the absolute value of the matching variable's standard deviation is less than 20% (26). Table 4 shows that, except for age and education, the standard bias of all variables after matching was within 20%, indicating that the matching result was credible. The visualization of the matching results is shown in Figure 2. Generally, using the PSM

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TABLE 4 Balance test for propensity score matching.

Variable	Unmatched	Mean		%bias	%reduc-tion	*t*-test	
	Matched	Treated	Control			*t*	*p* > *t*
Individual characteristics							
Hukou status	U	0.796	0.387	91.400		39.250	0.000
	M	0.796	0.798	−0.600	99.400	−0.290	0.769
Gender	U	0.561	0.554	1.400		0.590	0.555
	M	0.562	0.554	1.500	−9.900	0.710	0.475
Marital status	U	0.814	0.763	12.500		5.330	0.000
	M	0.814	0.769	11.000	12.100	5.140	0.000
Annual income	U	10.035	10.614	−62.100		−25.930	0.000
	M	10.045	10.012	3.600	94.200	1.550	0.122
Selection of medical and health services							
Hospitalization	U	0.116	0.099	5.400		2.260	0.024
	M	0.116	0.105	3.700	32.000	1.680	0.093
Medical expenditure	U	7.221	7.263	−2.700		−1.120	0.263
	M	7.220	7.187	2.100	20.800	0.970	0.334
Hospital scale	U	3.020	2.303	44.300		18.690	0.000
	M	3.020	3.059	−2.400	94.600	−1.100	0.273
Satisfaction with medical conditions	U	3.632	3.650	−2.300		−0.970	0.331
	M	3.632	3.628	0.500	79.700	0.210	0.830
Medical level	U	3.467	3.540	−8.400		−3.550	0.000
	M	3.467	3.500	−3.700	55.800	−1.700	0.090
Health behavior							
Smoke	U	0.563	0.610	−9.700		−4.100	0.000
	M	0.562	0.576	−2.800	71.500	−1.280	0.200
Drink	U	0.152	0.139	3.800		1.600	0.111
	M	0.152	0.149	0.900	77.500	0.390	0.696
Nap	U	0.557	0.610	−10.700		−4.490	0.000
	M	0.557	0.549	1.800	83.300	0.820	0.411

efficiently minimized discrepancies in numerous individual characteristics among the treated group and control groups before sample matching, and the treated group and control groups were more reliable.

3.3 Effect

The influence of URRMI on self-rated health of inhabitants was calculated using k-nearest neighbor matching. Table 5 shows the results of PSM-DID regression on randomly selected samples. Treated ×Time, hukou status, age, highest education, annual income, medical expenditure, hospital scale, clinic satisfaction, and napping were found to have an impact on the self-rated health of residents. The regression coefficient of the interaction term Treated ×Time was 0.456 and was statistically significant at the 10% level. This means that those who opted to engage in URRMI had a 0.456-unit greater coincidental in their self-rated health improvement compared to those who did not. Evidently, URRMI has a substantial impact on people's health.

Hukou status also had an impact on the self-rated health of residents. The regression coefficient of the PSM-DID was 0.311 and was significant at the statistical level of 5%, indicating that the self-rated health of rural hukou had a superior chance of ameliorating to urban hukou. Age also influenced the self-rated health of residents, and the regression coefficient for the self-rated health of residents was −0.046, which was significant at the statistical threshold of 1%, indicating that younger residents had the preferable probability of self-rated health. Contrary to traditional ideas, older individuals felt that their bodies were unhealthy. The regression coefficient for the variable highest education was 0.256 and statistically significant at the 1% level, showing that the higher the educational background, the better likelihood of the self-rated health. In contrast, individuals with a high educational background rated their health status as satisfactory. Annual income also had a significant impact on the self-rated health of residents at the 5% statistical level, with a regression coefficient of 0.162, indicating that

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TABLE 5 Propensity score matching and difference-in-difference result graph (Health).

Health	Coef.	Std. Err.	z	P > z	[95% Conf.]	[Interval]
Treated ×Time	0.456	0.257	1.770	0.076	−0.048	0.960
Time	−0.238	0.183	−1.300	0.195	−0.597	0.122
Treated	−0.252	0.183	−1.070	0.169	−0.611	0.106
Individual characteristics						
Hukou status	0.311	0.149	2.080	0.037	−0.604	−0.019
Age	−0.046	0.008	−6.020	0.000	0.031	0.062
Gender	0.036	0.194	0.190	0.852	−0.416	0.344
Marital status	−0.034	0.185	−0.180	0.853	−0.329	0.398
Highest education	0.256	0.069	3.680	0.000	−0.391	−0.120
Annual income	0.162	0.080	2.030	0.043	−0.318	−0.005
Selection of medical and health services						
Hospitalization	0.271	0.224	1.210	0.227	−0.710	0.168
Medical expenditure	−0.464	0.061	−7.650	0.000	0.345	0.583
Hospital scale	0.106	0.044	2.400	0.016	−0.193	−0.020
Clinic satisfaction	0.427	0.101	4.220	0.000	−0.626	−0.229
Hospital quality	0.050	0.090	0.560	0.577	−0.226	0.126
Health behavior						
Smoke	−0.077	0.191	−0.400	0.688	−0.298	0.452
Drink	0.295	0.200	1.480	0.140	−0.686	0.097
Nap	0.232	0.136	1.710	0.088	−0.498	0.035
_cons	2.918	1.038	2.540	0.005	−4.750	−0.611

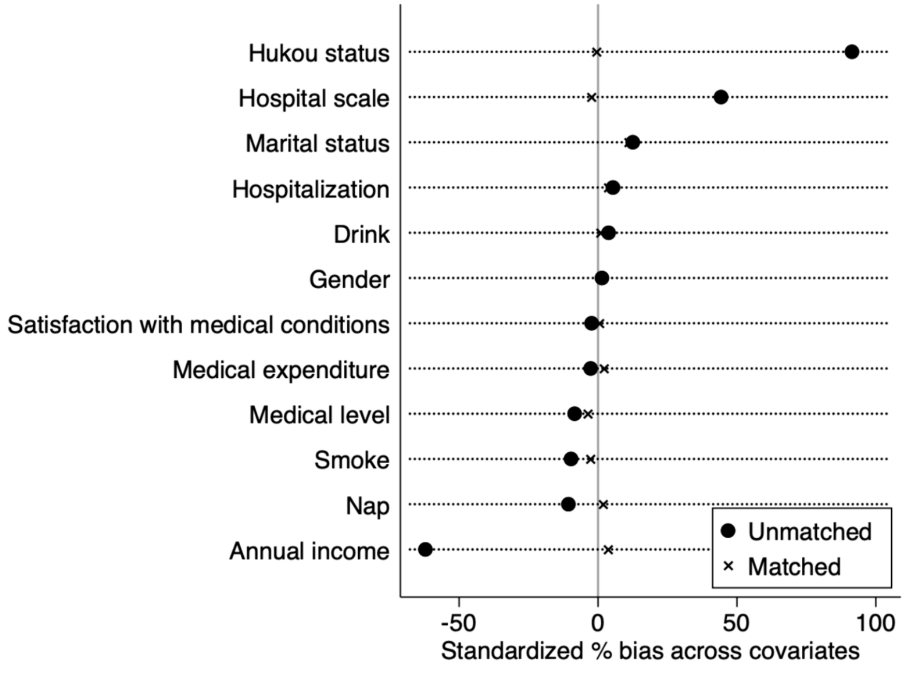

FIGURE 2
Balance test results of tendency propensity score matching score.

a higher annual income had more possibility for self-rated health and vice versa. Medical expenditure had a statistically significant effect on the self-rated health of residents at the 1% statistical level, and the regression coefficient was −0.464, indicating that payment of more medical expenses modified the chances of enhancing self-rated health while paying more medical expenditure worsened self-rated health. The Hospital scale statistically significantly affected residents' self-rated health at the 5% level. The regression coefficient was 0.106, indicating that individuals who go to major hospitals perceived that their health was better, but this observation may be a problem of two-way choice, which needs further research. In the CFPS questionnaire, medical conditions include medical equipment, drugs, medical quality, hospitalization conditions, the distance to seek medical treatment, and the convenience of transportation. Patients strived for advanced medical care at hospitals of different scales and provided their satisfaction ratings on the medical services they received. Through the statistics, we found clinic satisfaction had a statistically significant effect on the self-rated health of residents at the statistical level of 1%, and the regression coefficient was 0.427, indicating that residents who were satisfied with the hospital services were more likely to have the finest health condition. Nap had a significant influence self-rated health of residents at the statistical level of 10%. The regression coefficient was 0.232, indicating that individuals who has lunch break habits were more potential to possess favorable self-rated health. The study found that the URRMI had a significant effect on improving the health status of insured individuals. Insured individuals had a higher probability of having better health status compared to those who did not purchase insurance. Younger individuals, those with higher education levels, those with higher average annual income, those who spent less on medical expenses, larger scale of medical treatment, and higher satisfaction with medical treatment, those who has a nap habit had a higher probability of improvement in health status.

3.4 Further exploration of URRMI and medical expenditure

To delve deeper into the factors that influence URRMI in objective health indicators, we have devised a novel model that examines the correlation between URRMI and medical expenses. This model offers a more thorough understanding of the interplay between URRMI and self-perceived health status of individuals. Specifically, we have introduced a new dependent variable, expense. This variable is assigned a value of 1 if the Medical expenditure of a given sample constitutes less than 5% of its annual income; otherwise, it is assigned a value of 0. As for the explanatory variables and covariates, we have eliminated the original Medical expenditure variable while retaining all other variables unchanged.

In conducting this analysis, we continue to utilize the PSM-DID model that was previously mentioned, as it remains a suitable framework for examining the relationships and effects in question. The model settings are shown in Equation 3:

$$logit\left(Treated_i = 1\right) = \gamma_2 + \gamma_3 X_i + \varepsilon_{it} \tag{3}$$

where i represents the individual. *Treated* indicated participation in URRMI and was assigned a value of 1 if the individual participated and 0 otherwise. X_i is the matching variable, which included Hukou status, Gender, Marital status, Annual income, Hospitalization, Hospital scale, Clinic satisfaction, Hospital quality, Smoke, Drink, and Nap. ε_{it} expresses error terms and contains information other than the main variables of the model.

The following equation of the DID model was estimated for continuous outcomes:

$$expense_{it} = \alpha_4 + \alpha_5 Treated_{it} + \alpha_6 Time_{it} + \alpha_7\left(Treated_{it} \times Time_{it}\right) + \beta_i \times X_{it} + \varepsilon_{it} \tag{4}$$

Where $expense_{it}$ stands for the expenditure of medical i at time t, with Medical expenditure ≤ 5%Annual income = 1, Medical expenditure >5%Annual income = 0. The treated variable $Treated_{it}$ is a binary indicator; it epitomizes the group dummy variable. $Treated_{it} = 1$ indicates that an individual i belongs to the treated group, having participated in the URRMI. $Treated_{it} = 0$ epitomizes the control group, indicating that the individual i did not participate in the URRMI. $Time_{it}$ epitomizes the time dummy variable, with $Time_{it} = 0$ indicating the time t before the individual i participated in the URRMI (the year 2018) and $Time_{it} = 1$ indicating the time t after the individual i participated in the URRM (the year 2020). The variable $Treated_{it} \times Time_{it}$ signifies the collaboration among groups and time. X_{it} epitomizes a set of individual covariates of individual i at time t. ε_{it} expresses error terms and contains information other than the main variables of the model.

Table 6 presents the results of a PSM-DID analysis of the dependent variable, expense, and its independent variables. We can observe that the coefficient for the key variable Treated × Time is −0.357, which is significant at the 5% level. This indicates that for the treated group who purchased insurance, their likelihood of incurring higher medical expenses has increased. Compared to the control group who did not purchase insurance, the treated group who did purchase insurance has a higher probability of spending more on medical expenses. This may suggest that the insurance purchase behavior has, to some extent, increased the utilization of medical insurance. This is also the outcome we anticipated.

3.5 Testing the robustness of PSM-Did estimation

In this study, a placebo test was used as the robustness verification method. The specific implementation method involved randomly selecting the same number of samples as the original control group as the dummy control group from the samples. The remaining samples were then designated as the dummy experimental group. The regression parameters were re-estimated 500 times under the condition, keeping the relevant control variables, matching methods, and regression steps unchanged.

A placebo test was used to validate the reliability of the PSM-DID approach for estimating the core explanatory variable α_3. Figure 3 depicts the distribution of estimated coefficients for the interaction term after 500 trials, with the estimated coefficients shown. Close to the ordinary normal distribution, α_3 was evenly dispersed about 0. The position of coefficient α_3 (0.337) is indicated by the vertical bar

TABLE 6 Propensity score matching and difference-in-difference result graph (expense).

Expense	Coef.	Std. Err.	z	P>z	[95% Conf.]	[Interval]
Treated ×Time	−0.357	0.155	−2.31	0.021	−0.660	−0.054
Time	0.154	0.121	1.27	0.205	−0.084	0.392
Treated	0.184	0.116	1.59	0.113	−0.043	0.410
Individual characteristics						
Hukou status	0.096	0.097	0.99	0.321	−0.094	0.286
Age	−0.026	0.004	−6.62	0.000	−0.034	−0.018
Gender	0.507	0.119	4.28	0.000	0.275	0.740
Marital status	−0.060	0.108	−0.55	0.580	−0.272	0.152
Highest education	0.238	0.041	5.85	0.000	0.158	0.318
Annual income	1.242	0.063	19.79	0.000	1.119	1.364
Selection of medical and health services						
Hospitalization	−4.031	0.199	−20.30	0.000	−4.420	−3.642
Hospital scale	0.327	0.027	12.10	0.000	0.274	0.380
Clinic satisfaction	0.173	0.058	2.99	0.003	0.060	0.286
Hospital quality	−0.136	0.053	−2.53	0.011	−0.240	−0.031
Health behavior						
Smoke	0.024	0.119	0.21	0.837	−0.209	0.257
Drink	0.244	0.119	2.05	0.041	0.010	0.478
Nap	0.089	0.079	1.13	0.259	−0.065	0.243
_cons	−12.005	0.696	−17.25	0.000	−13.370	−10.641

(Figure 3). This number emerges at the end of the kernel density distribution plot, demonstrating that basic medical insurance does not significantly improve the health of insured residents under the URRMI scheme once bogus experiments are constructed. This thus lends credence to the preceding conclusions.

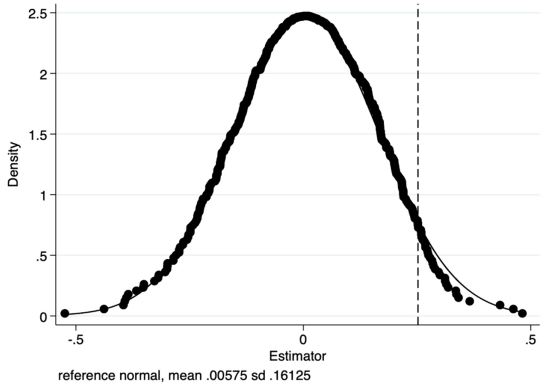

FIGURE 3
K-Nearest neighbor matching the distribution of 500 random sampling regression coefficients.

4 Discussion

The PSM-DID methodology effectively combines the strengths of PSM and DID analysis. Specifically, PSM aids in selecting control group who possess similar characteristics to the treated group, thus mitigating the potential for endogenous self-selection bias. Conversely, DID serves to precisely quantify the impact of the URRMI policy by comparing the changes observed in the treated group with those in the control group, both before and after the implementation of the URRMI policy. There is ample evidence from numerous journal articles to support that this model is widely utilized in the medical domain as well as in other disciplines due to its robust analytical framework. A study adopted PSM-DID to evaluate whether the aggregated payment based on cases of acute patients and chronic patients is more cost-effective than the traditional payment of a daily allowance for acute and chronic patients (27). A Japanese study examined the spillover consequence of the Japanese public long-term care insurance (LTCI) as a policy to encourage labor force participation of family caregivers from 1995 to 2013 (28). The study examined the spillover consequence in two periods: before and after the LTCI's introduction in 2000 and before and after its major amendment in 2006 (28). A study from China used PSM-DID to assess the impact of the URRMI scheme on how often urban and rural residents in the four experimental areas used medical services (29). Using panel data, a different study used PSM-DID to examine the targeting and impact of the Askeskin program, a sponsored social health insurance program for the underprivileged and those living in poverty. The study showed that the program targeted the underprivileged and those who are most at risk of catastrophic out-of-pocket medical expenses (30). A study used PSM-DID to assess the impact of the restructurings on the two most prevalent childhood infections, the incidence and treatment of fever (malaria) and diarrhea infections, after introducing the national health insurance scheme restructurings (31). Notably, previous

evaluation studies have not examined these outcomes after introducing the national health insurance scheme reforms (31). To assess the impact of China's National Essential Medicine Policy (NEMP) on outpatient treatment utilization and spending in Tianjin, China (32). Accordingly, the utilization of this robust PSM-DID methodology in this study not only adeptly mitigates the challenges posed by endogenous self-selection bias but also offers a rigorous analysis and insightful discussion of the causal relationship between the URRMI policy and self-rated health outcomes.

This study investigated the impact of URRMI on the health of residents and showed that URRMI can greatly improve the health of insured people. Like most prior conclusions, this study indicates that URRMI has a considerably favorable impact on the health of inhabitants. This could be because enrolling in health insurance can affect older persons' decisions to seek medical care, thereby dramatically reducing healthcare demand (HCD) and healthcare expenditure (HCE) (33). URRMI reduces out-of-pocket expenditure for residents, relieves the financial burden of medical expenses, and lowers residents' medical costs. It also gives people a sense of security and solves the most fundamental health and livelihood issues. According to some studies, there is a time lag between the recovery and improvement of residents' health levels after enrolling in basic medical insurance. This implies that the change in residents' health levels caused by the accumulation of health stock is quantitative to the qualitative change process. This makes measuring the influence of URRMI on residents' health at the start of implementation or over a short period difficult (34). The participation rate of basic medical insurance in China has remained above 95% throughout the year, but it has not achieved full coverage. Referring to the reform of the medical insurance system in Germany and Japan, the main way to quickly realize universal medical insurance is to force residents to participate in insurance by legislation. Therefore, the modification of URRMI in China needs to be coordinated with the corresponding laws and regulations to further improve the coverage of the scheme. In this way, the law can be a factor in amplifying the health performance of basic medical insurance.

Compared with objective health indicators, choosing self-rated health status as the dependent variable of this study has many advantages. Firstly, it reflects individuals' subjective perception of their own health status, which can better reflect individuals' true feelings than objective indicators. Secondly, self-rated health status is easy to collect and does not require complex medical examinations or biological indicator measurements, reducing research costs and difficulties. Lastly, self-rated health status reflects non-biological factors such as individuals' psychological state and lifestyle, which often have important impacts on health status and can be examined through self-rated health status. However, self-rated health status is prone to subjective factors, and its evaluation may differ among individuals based on their psychological state, social and cultural background, and health knowledge level. Additionally, it does not reflect biological information that objective indicators, such as blood pressure and blood sugar, can indicate, which are important for evaluating the health status of the individual.

We also found that the geographic difference between urban and rural hukou has a significant effect on the health of residents. Our statistical results show that rural hukou had better self-rated health status. Firstly, the household registration system is a unique administrative system of identity management in China. The state lawfully collects, confirms, and registers basic information such as birth, death, kinship, and address of residents, to safeguard the rights and interests of some residents in employment, education, social welfare, and other aspects. Therefore, it has also formed a dual structure of urban and rural areas specific to the context of China. As the results illustration, the characteristics presented by urban hukou and rural hukou are noticeably distinctive. Additionally, Fang H et al. emphasized in their article that urban inhabitants consistently revelation poorer health outcomes compared to rural residents, with a sensitive risk of being diagnosed with critical illnesses (35). The unambiguous contrast in the natural environment and lifestyle between urban and rural zones is noteworthy. The rural environment is relatively fresher, without continuous exhaust pollution, light pollution, and noise pollution. Despite the ongoing efforts to enhance the construction of green areas in urban area, it is incontrovertible that rural areas possess a more abundant presence of green spaces. Notably, a study has authenticated the momentous role of green spaces in optimistically enhancing self-perceived general health and mental well-being, thus corroborating our findings (36). Thirdly, the gradual expansion of medical resources into rural zones has led to substantial improvements in the rural healthcare system and its resources (37). This has translated into a significant enhancement in basic medical service capabilities, surpassing previous standards. The rural health education and promotion initiatives are being undertaken on an extensive scale. Through the placement of health promotion posters and the organization of public welfare lectures at village activity centers, the health literacy and healthy behavior patterns of rural residents have been strengthened. Such endeavors are instrumental in preventing and managing the prevalence of various chronic diseases. However, the adjustment of urban and rural medical insurance to urban resident medical insurance has changed the medical insurance benefits, which helps to narrow the urban–rural gap to some extent. After combating poverty, the government has also implemented multiple policies to ensure the effective linkage between consolidating and expanding the achievements of poverty alleviation and promoting rural revitalization. The reform of the medical insurance system will be continuously intensified to ensure basic medical security for low-income rural residents and firmly prevent a large-scale return to poverty caused by illnesses. Further research and continuous monitoring may be needed to assess the impact of the changes caused by the deepening reform of the medical security system.

Given the differences among urban and rural zones, the reform path of URRMI can start from the following three aspects. First, the government must persist in enhancing the rural grass-roots medical system. It is highly recommended that the government annually augment its financial investment in rural grass-roots medical services and facilities by a designated percentage, thereby guaranteeing a steady increase in medical resources in rural areas. Regarding resource allocation, the government must effectively ensure the efficient and rational utilization of resources, tailored to the specific needs and population structure of each region. Additionally, incentives should be offered to medical graduates to work in grass-roots medical institutions, accompanied by commensurate policy support and welfare benefits. Second, the government should further intensify the reform of the URRMI payment plan and promote a diversified composite medical insurance payment method (38), aiming to better accommodate the diverse characteristics of medical services. Third, improve the multi-level construction process of URRMI systems, divide rural

areas into different medical insurance access standards through financial subsidies, and regulate the process of social wealth redistribution. Fourth, the reform of the URRMI should align with the government's macro-control measures on the medical and health service market to improve the influence of purchasing with quantity on the prices of medical drugs and devices. Fifth, government agencies encompassing health, education, and culture ought to intensify their interdepartmental collaboration and harness the power of diverse social media platforms to disseminate information on health literacy and policies pertaining to basic medical insurance for both urban and rural residents, ultimately fostering a deeper understanding and awareness among the populace.

Contrary to previous research findings, our study presents a novel discovery: traditional health behaviors such as smoking, alcohol consumption, and napping do not always have absolute and unchanging effects on health. Specifically, our data indicates that simply controlling smoking and drinking habits does not significantly improve health outcomes. However, it is noteworthy that we found that appropriate napping has shown significant positive impacts on health and regression analysis, strongly suggesting that individuals with a habit of napping tend to have better health conditions. This conclusion aligns with numerous mainstream studies. Among various research reports, one study particularly caught our attention. It suggests that brief afternoon naps indeed have a positive effect on promoting human health (39). This discovery further reinforces our view that good health habits are of indispensable importance for maintaining individual health status.

After conducting a further exploration of URRMI and Medical expenditure, we discerned a notable rise in the probability of the experimental group encountering steeper medical costs compared to other groups. Moreover, our analysis of self-rated health levels revealed a compelling pattern: as medical expenses escalated among the sampled individuals, their self-assessed health status showed a concurrent decline. These findings align closely with previous research, indicating a correlation between increased health service utilization and a decline in self-rated health (40). This pattern suggests that medical insurance purchases, to a degree, have encouraged the utilization of medical insurance benefits, yet it is also intertwined with elevated medical necessities and health challenges. Evidently, URRMI shifts the burden of personal medical expenses onto the insurance system, prompting insured individuals to seek out specialized medical services and facilities whenever illness strikes. This augmentation in healthcare utilization is inevitably mirrored in the utilization of medical insurance. Nevertheless, this trend may also stem from heightened medical needs and health issues. Those who frequently avail themselves of healthcare services often rate their health status lower, likely due to the more significant health problems they encounter, necessitating more extensive medical interventions. Consequently, when delving into the intricate relationship between medical insurance purchases, medical expenditures, and self-rated health, we must adopt a multifaceted approach, considering various factors. Future research can delve deeper into the interplay between these variables and explore ways to refine medical insurance policies to enhance overall health standards and quality of life.

5 Limitations

This study had respective limitations. First, a significant assumption of PSM-DID regression is that the model should contain all the covariates before and after the match that may affect the effectiveness of the strategy. Unobservable covariates will cause divergent tendencies between treated groups and control groups, which may bias the results of this study. Second, PSM is for section data, and DID is for panel data. Generally, there are two solutions to solve the problem of different application ranges. Panel data can be converted into section data for processing, and then phase-by-phase matching is conducted on each phase section of panel data. This study adopted the phase-by-phase matching method of a balanced panel. Although this method can solve the problem of sample matching in different periods, special class variables might still cause sample mismatch (41). Finally, the survey relied on specific time points and did not continue to study and compare data before 2018. Consequently, the data from the two periods are not significant in the short term, and the long-term effects of policies are ignored.

6 Conclusion

This study further confirms that PSM-DID is an imperative tool for scrutinizing the influence of the amalgamation of URRMI on self-rated health status through 2 years of panel data. Moreover, the statistics imply that purchasing in URRMI has an optimistic impact on the health of residents, promoting better self-rated health performance. In addition, we also institute regional differences in health, with urban residents having superior health status compared to rural residents. Furthermore, our study challenges previous research, revealing that traditional health behaviors have varying effects, with smoking and drinking control not significantly improving health, while appropriate napping positively impacts health, aligning with mainstream studies and emphasizing the importance of good health habits. Our further exploration of URRMI and medical expenditure revealed a rise in medical costs for the experimental group and a correlation between increasing expenses and declining self-rated health, aligning with research showing a link between increased health service use and poorer self-rated health.

We recommend that the government expand the coverage of the URRMI scheme, narrow the gap among urban and rural zones, and improve the implementation of certain insurance subsidies and benefits for rural hukou to improve the overall health level of residents. Furthermore, the government can support medical institutions to provide better and more affordable medical facilities for urban and rural residents by raising the standard and scope of medical insurance payments. Finally, policymakers should learn from the implementation of the experimental areas, improve the laws and regulations related to URRMI, establish a sound medical insurance supervision mechanism, strengthen the administration and supervision of medical insurance funds, and formulate more efficient implementation strategies for the forthcoming nationwide promotion.

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

YS-Y: Conceptualization, Data curation, Formal analysis,Investigation, Methodology, Writing – original draft, Writing – review & editing. CY-T: Writing – review & editing. XX-Y: Writing – review & editing. WD: Writing – review & editing. LX-H: Writing – review & editing. LW: Writing – review & editing. PT: Writing –review & editing. MX-H: Conceptualization, 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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Restricted health service utilization and subsequent positive self-care behavior during the early COVID-19 pandemic in China

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Background: The reallocation of health resources, epidemic prevention and control measures during the COVID-19 pandemic triggered widespread restricted health service utilization, some residents and patients tried positive self-care behavior to maintain their health, yet the efficacy of this intervention remains unclear.

Object: Based on the reasoned action approach (RAA) theory, this study aimed to investigate the correlation between self-care behavior and restricted health service utilization among adults in China, trying to discover the vulnerable groups and external and intrinsic factors that affect self-care behavior among Chinese adults.

Methods: Data on demographics, socioeconomic, health status, and self-care behavior were collected in "The Early China COVID-19 Survey," a cross-sectional anonymous online survey of the general population in China. Self-care behavior was measured by four indicators: weight control (WC), physical activity (PA), prevention behavior (PB), and online medical consultation (OMC). The multiple linear models and binary logistic regression were used to examine whether restricted health service utilization (RHSU) is associated with self-care behaviors; also, adjusted multivariate logistic regression was used to analyze subgroup heterogeneity.

Results: In total, 8,428 adult participants completed the survey, the mean OMC score was 1.51 (SD 1.34), the mean PB score was 18.17 (SD 3.44), and the proportion of participants who engaged in WC and PA was 42.30 and 62.57%, respectively. According to the multiple regression model, the RHSU was significantly positively correlated with all four indicators of self-care (WC: OR = 1.34, $p < 0.001$, PA: OR = 1.34, $p < 0.05$, MC: OR = 1.30, $p < 0.001$, PB: coef = 0.16, $p < 0.05$). We also observed some significant differences in the intensity of this relationship by subgroup analysis, precisely, OMC (high vs. moderate vs. low infection-risk level: OR = 1.48; 1.41; 1.19, $p < 0.1$), PA (male vs. female: OR = 1.27;1.06; $p < 0.05$, high vs. Moderate and low infection-risk level: OR = 1.51; 1.17; 1.02, $p < 0.05$), PB (Chronic disease groups vs. no: coef = 0.46; 0.1, $p < 0.05$).

Conclusion: Restricted health service utilization predicts more positive self-care behavior, and the intensity of partial correlation was significantly different in the subgroups of sex, actual infection risk level of the living area, and chronic diseases. These findings highlight the urgent demand for self-care behavior

among Chinese adults during the pandemic and provide new insights for developing self-care and reducing the burden on the healthcare system in the long term.

KEYWORDS

health service utilization, self-care behavior, reasoned action approach, online medical consultation, subgroups analysis, vulnerable population, COVID-19

Introduction

During the first wave of the COVID-19 pandemic, a global lockdown was imposed in response to the rapid spread of the virus; the government's regulations or restriction measures to curtail virus spread may have an enormous impact on people's daily lives (1, 2). It is self-evident that the implementation of quarantine measures, lockdown, and social distancing protocols also led to some negative repercussions, which include a significant meltdown in economic growth, an increase in unemployment or financial insecurity, a rise in the cost of living, and a severe impact on health service utilization among the population (3–5).

With the continued containment and control of the COVID-19 pandemic, the general population also experienced unintended potential health risks arising from the restriction of routine healthcare services, specifically, the consequences of the pandemic for the health of these non-COVID-19 patients, including delayed timely detection and treatment, avoided and delayed emergency department visits, unmet healthcare needs and increased rates of exacerbation as resources were reallocated to urgent care for COVID-19 patients (6), researches noted the COVID-19 pandemic has adversely affected the healthcare utilization of the population worldwide, and situation of restricted health service utilization is alarming (7, 8). A national longitudinal study from China found the most considerable negative impact of the COVID-19 pandemic on health services utilization was observed between Jan 2020 and Apr 2020, with approximately a 32% reduction in hospitals, a 22% reduction in community health centers, and 27% reduction in township centers (9). Studies from Europe have claimed that during the COVID-19 pandemic, individuals with acute myocardial infarction, stroke, heart failure, and other chronic cardiovascular diseases experienced a significant reduction (40%) in hospitalization rates and emergency department visits compared to baseline (10). Globally, a large study indicated that global healthcare utilization showed a median decline of 37.2% between the pandemic and pre-pandemic periods, ranging from 19.8 to 50.5% (8). In short, the fear of infection and reduced availability of medical services have driven down non-COVID-19 healthcare utilization. The demand of the general population, particularly vulnerable groups like chronic patients, to improve their health status is a difficult task under normal circumstances, the obvious barriers imposed by the COVID-19 pandemic have considerably increased this difficulty. Meanwhile, considering the healthcare system supply is limited, it appears to be a moral dilemma between allocating medical resources to contain the spread of the virus or providing adequate healthcare service during the pandemic. Nevertheless, it also indirectly drives us to be more concerned about exploring strategies and measures to address the restricted health service utilization more effectively.

Background

Since the outbreak of the COVID-19 pandemic, the infection spread has come in waves in China, restrictions on health services were occasional (9), and growing research has focused on self-care behavior and recommended it as a response strategy to tackle the potential health harms arising from restricted health service utilization (11, 12). Drawing on the various relevant literature, the restricted health service utilization (RHSU) not only hinders surveillance of health status but also will likely lead to some worse outcomes if it continues, such as unhealthy lifestyles, treatment interruption, and worsening of chronic disease symptoms (13), all of which could lead to serious health problems. To alleviate this potential health crisis, the people with intentions to maintain health and those with chronic diseases preferred to adopt non-therapeutic measures, including physical activities, dietary habits, self-care monitoring and online medical consultation after the outbreak (14–16). Previous studies found that several types of health service utilization restrictions were associated with self-care activities, such as both primary and specialized care (17), personal care aides (18), primary mental health care service (19) and chronic disease treatment (20, 21). Drawing upon evidence from related research, the RHSU might directly or indirectly motivate the general population to compromise in daily choices like consumption habits, diet, exercise, self-care activity and medical consultation, and most people had mitigated potential health harms through these behavioral changes (14–16, 22). For instance, a study of stroke survivors stated that self-care behavior such as physical activity, diet, weight control, smoking cessation, and abstinence from alcohol could help those with restricted health services to reduce systolic and diastolic blood pressure and decrease the occurrence of complications such as hyperglycemia and diabetes and keep disease stable (23). Medical practitioners have also suggested that adopting self-care activities and improved self-care behaviors may play a critical role in maintaining health or preventing immediate and subsequent complications during the COVID-19 pandemic (24).

Specifically, self-care behavior is recognized as an essential and valuable behavior because it emphasizes the positive role of people in maintaining their health, and common individual self-care activities include engaging to improve health status, prevent disease, limiting illness, and regain health (25), the beneficial effects of self-care include improved well-being and lower morbidity, mortality, and healthcare costs (21). Previous research has revealed the complexity of self-care

Abbreviations: RAA, Reasoned action approach; RHSU, Restricted health service utilization; COVID-19, Coronavirus disease 2019; WC, Weight control; PA, Physical activity; PB, Prevention behavior; OMC, Online medical consultation.

and illustrated the wide variety of factors that influence the decisions individuals make about engaging in self-care (13–16); it also investigated the difficulty performing self-care among special populations (e.g., multiple chronic conditions, severe mental illness, low health literacy) (26); and sustained evolving technology to enable individuals to manage their conditions and improve the efficiency of self-care (27), the multiple disciplines are actively studying self-care and contributing variable knowledge to the topic nowadays (28). However, the current literature suggests that self-care is still under-appreciated and insufficiently understood; there are many challenges to the prevalence of self-care behavior (26). Thus, there is a need to explore the mechanisms underlying self-care behavior further to drive relevant policy development, especially for those countries and regions with a growing burden on health service systems.

Research gaps and the present study

Previous studies have been conducted on self-care behavior in the older adult, chronic disease patients, non-communicable disease patients, and healthcare professionals (14–16, 28). However, there is still a lack of continuity across research initiatives, and few studies have investigated the changes in self-care behavior among whole adult populations during the pandemic. In addition, some research noted that demographic, socioeconomic status, and health status determine the acceptance and engagement of self-care behavior interventions (26, 29). However, the direct evidence for the relationship between the RHSU and self-care behavior is limited and controversial, and the discussion about the possible differential impacts of RHSU on self-care behavior of individuals with varying health status may differ is also not well substantiated. These knowledge gaps may lead to a deficiency in the comprehensive understanding of the underlying mechanisms of self-care behavior in the current Chinese population and diminish the identification of vulnerable groups with worse self-care behavior. In addition, although some studies have supported the viability of self-care behavior as a strategy to address the RHSU problem and reduce the burden on the healthcare systems, the direct evidences are limited and controversial.

Based on the literature review, we found that the instruments used to measure self-care behavior consisted of several main aspects, such as health consciousness, nutrition, physical activity, sleep quality, medical management of the disease, seeking social support, and adherence to the recommended regimen (30–32), however, previous studies have commonly used single or one-dimensional indicators to measure self-care behavior. Therefore, we decided to establish indicators to measure self-care behavior across two main dimensions, including autonomous behavior and consultative behavior (33). Autonomous behaviors are implemented directly by the patient, such as changing activity or taking weight control to make the symptoms decrease or go away and adjusting health habits to avoid infections. Consulting behaviors are based on guidance from healthcare providers; for example, call your healthcare provider for advice and seek health consultation by using other channels. Additionally, the COVID-19 pandemic provides a particular perspective on whether self-care behavior could be considered an effective strategy for mitigating the health risks caused by restricted health service utilization. Since the reason for the impact of this restricted health service utilization is mainly exogenous and individuals receiving routine medical care or medical consultations also experienced health risk shocks caused by the COVID-19 pandemic. Furthermore, previous research on the relationship between health service utilization and health behavior has occasionally been controversial (18, 20); for example, when experiencing restricted health service utilization, some residents and patients may prefer to receive health service elsewhere rather than engage in more positive self-care behavior. However, the COVID-19 pandemic significantly affected nationwide health service utilization (9), which indirectly helps us to rule out some confounding factors.

According to the reasoned action approach (RAA), a common theory used to understand health behaviors (34), we assume that every individual has the willingness to maintain health and confidence to control their health behavior, also fully aware of the consequences of RHSU and the norms of self-care behavior, then we could assume RHSU is targeted for interventions to change self-care behavior. In our study, we expect that adults who experienced RHSU would be more likely to report positive self-care behavior to maintain their health. However, more evidence is required to explore differences in risk perceptions and self-care behavior at the individual level.

Overall, in this study, we aimed to explore the following question based on a large nationally representative sample:(1) the relation between restricted health service utilization (RHSU) and self-care behavior among Chinese adults during the COVID-19 pandemic; (2) whether the relationship between self-care behavior and restricted health service utilization (RHSU) was different across social groups (i.e., male, female, 18–44 age, 45 and above age; chronic diseases adults, no-chronic diseases adults; adults living in the low, moderate and high infection-risk level area).

Methods

Sample collection

Data on demographics, socioeconomic, health status and self-care behavior were obtained from an anonymous online survey called the "2020 China COVID-19 Survey," which was collected between late April and mid-May 2020. It was collected via WeChat, which is a popular social media tool that has become an essential part of the daily work and life of Chinese adults. The primary aim of the 2020 China COVID-19 Survey study is to explore whether health disparities by age, sex, race, living condition, or socioeconomic status emerge or worsen throughout the pandemic; this survey has been used in other articles in China during the COVID-19 pandemic (35, 36). This structured questionnaire compasses seven topics:

1 The demographic and socioeconomic characteristics.
2 Health status, including chronic diseases during the COVID-19 pandemic.
3 Awareness, attitude, knowledge, and practices toward COVID-19.
4 COVID-19 experiences and impacts.
5 Medical consultation habits.
6 Behaviors that could prevent the spread of COVID-19.
7 Lifestyle habits.

To ensure data accuracy and integrity, one project manager was recruited in each province to coordinate the province-wide survey and

organize survey training, and six to twelve local investigators were recruited in each city based on household incomes to distribute online questionnaires and control the survey quality. After being trained in online data collection, each local investigator was asked to send the online questionnaire directly to 20–30 local households in their social networks, including relatives, neighbors, friends, and workmates. Each eligible participant was invited to complete the online questionnaire, which they completed in an average of 8.5 min. Participants are given an appropriate gift when they complete the questionnaire. Some of the older adult could not participate in the online survey due to their age and education level; regarding this group, we decided that relatives living with them obtain their answers through oral questioning and fill out the survey based on their options (26). When this survey began, the COVID-19 pandemic had already caused more than 83,000 infections in mainland China and over 4,600 deaths (37). The pandemic was generally disseminated, with clusters of outbreaks caused by transmitted cases occurring in some areas. Since April 2020, the corresponding preventive and control measures in most provinces in China have been downgraded from emergency response to regular management, with social quarantines, blockades, and travel restrictions identified and implemented according to regional risk classifications. In this study, we used targeted stratified convenience sampling to select residents in China's eastern, central, and western regions. Our survey included 8,428 adults aged 18 years and over 31 provinces in China. The survey was completed voluntarily and anonymously, and because of the high standardized quality control of the questionnaires, the baseline survey response rate is good. All subjects gave informed consent before participating in the survey, and the protocol was approved by the Ethics Committee Committee of Xi'an Jiaotong University (No. 2020–1,172).

Outcome variables

In our study, we used four indices, including weight control, physical activity, prevention behavior, and online medical consultation, to measure self-care behavior during the COVID-19 pandemic as the outcome variable in our analyses. Specifically, Weight control and physical activity are two typical variables in autonomous behavior dimensions; we dichotomized both variables to indicate if participants engaged in weight control or physical activity (Yes/No) during the COVID-19 pandemic. Prevention behavior was assessed using a scale consisting of four items derived from some questionnaires widely used to assess the disease control and prevention of populations during the outbreak of COVID-19 (3, 38, 39); the participants were asked about how often they had practiced the following preventive action: (1) Wear face mask in public settings, (2) Wash your hands after a trip outside, (3) Avoid unnecessary outings as much as possible, (4) avoid gathering as much as possible. Each item was rated on a 5-point Likert scale, and response options included: Never = 1, Rarely = 2, Occasionally = 3, Sometimes = 4, All the time = 5. Total scores ranged from 4 to 20, and higher scores mean better Prevention behavior. The Cronbach's alpha of this scale was 0.949. Regarding online medical consultation, participants were asked if they had used the following approaches to seek medical consultation during the COVID-19 pandemic; option approaches include online consultation, video consultation, telephone consultation service, and mail-order or personal delivery pharmacy. In this case, each option counts for 1 point, and the total score ranges from 0 to 4. Higher scores indicate that participants had used more approaches to seek medical consultation, and Cronbach's alpha of this scale was 0.782. To differentiate medical consultation capacity from individuals, based on the average of participants' response scores, we defined participants who chose two or more of the four options as "above-average performance in medical consultations." Thus, Online Medical behavior can be regarded as a binary variable (No = below average, Yes = above average).

Independent variable

RHSU was the independent variable, measured by recording whether the participant's routine medical care or medical consultations were restricted due to the COVID-19 pandemic. Hence, it was a binary variable (No = unrestricted due to COVID-19; Yes = restricted due to COVID-19).

Control variables

The control variable in our model includes demographic and socioeconomic characteristics mainly including region (city/rural or town), sex (male /female), age (18–44/45 years or above), education level (obtaining a bachelor's degree/no bachelor's degree), marital status (married/unmarried or divorced or widowed), the household income gradient in the last year (i.e., before the pandemic) was divided into three tertiles (low = 1st tertile/middle = 2nd tertile/ high = 3rd tertile), health conditions included chronic medical conditions (yes/no), self-rated health status (fair or poor/good/very good) and actual COVID-19 infection risk level (low/moderate/high) in the participant's place of residence. In addition, this study collected six variables on individual perceptions and impacts during the covid-19 pandemic, including whether participants think one of their family members had been infected with COVID-19 (yes/no), whether they had experienced food or medicine shortages (yes/no), whether they or their family members lose their job due to COVID-19 (yes/no), the degree of difficulty your family experiences in daily activities caused by COVID-19 related financial strain (no difficulty at all /mild difficulties/extreme difficulties), and the degree of how serious of a public health threat they think COVID-19 is or might become (low/midden/high).

Statistical analysis

From the survey, we collated the required descriptive statistics, including frequencies (*N*) and percentages (%) or means (M) and standard deviation (SD) and their 95% confidence intervals (95%CI). Then, we use multiple linear models and binary logistic regression to examine whether RHSU was associated with self-care behavior outcomes. In the process, we measured self-care behavior through four indicators and introduced the following control variable into the model for each indicator: demographic and socioeconomic variables (age, sex, marital status, educational level, residential area, and household income level in the last year), health condition (chronic disease, self-rated health) and COVID-19 related variables (lost job due to COVID-19, food shortage, experienced COVID-19 infection,

TABLE 1 General characteristics of participants [*N* (%)].

Variables		Total (*N* = 8,428)	Restricted health service utilization		*P* value
			No (*N* = 4,450)	Yes (*N* = 3,978)	
Region	City	5,085 (60.33)	2,516 (56.54)	2,569 (64.58)	< 0.001
	Rural	1,276 (15.14)	685 (15.39)	591 (14.86)	
	Town	2067 (24.53)	1,249 (28.07)	818 (20.56)	
Actual infection risk level of living area	Low	3,787 (44.93)	2,310 (51.91)	1,477 (37.13)	< 0.001
	Moderate	3,065 (36.37)	1,627 (36.56)	1,438 (36.15)	
	High	1,576 (18.70)	513 (11.53)	1,063 (26.72)	
Married status	Unmarried/divorced/ widowed	2,952 (35.02)	1,477 (49.97)	1,477 (50.03)	< 0.001
	Married	5,476 (64.97)	2,978 (54.33)	2,501 (45.67)	
Sex	Male	3,694 (43.83)	1761 (39.57)	1933 (48.59)	< 0.001
	Female	4,734 (56.17)	2,689 (60.43)	2045 (51.41)	
Age (years)	18–44	7,387 (87.65)	3,883 (87.26)	3,504 (88.08)	< 0.001
	≧45	1,041 (12.35)	567 (12.74)	474 (11.92)	
Bachelor degree	No	3,628 (43.05)	1952 (43.87)	1,676 (42.13)	0.109
	Yes	4,800 (56.95)	2,498 (56.13)	2,302 (57.87)	
Household income level	Low	3,587 (42.56)	1931 (43.39)	1,656 (41.63)	0.024
	Medium	2,149 (25.50)	1,156 (25.98)	993 (24.96)	
	High	2,692 (31.94)	1,363 (30.63)	1,329 (33.41)	
Self- rated health status	Fair or poor	233 (2.76)	91 (2.04)	142 (3.57)	< 0.001
	Good	1,336 (15.85)	617 (13.87)	719 (18.07)	
	Very good	6,859 (81.39)	3,742 (84.09)	3,117 (78.36)	
Chronic disease	No	6,745 (80.03)	3,759 (84.47)	2,986 (75.06)	< 0.001
	Yes	1,683 (19.97)	691 (15.53)	992 (24.94)	
COVID-19 infection (participants or family member)	No	7,643 (90.69)	4,299 (96.61)	3,344 (84.06)	< 0.001
	Yes	785 (9.31)	151 (3.39)	634 (15.94)	
Lost job due to covid-19	No	5,478 (65.00)	3,429 (77.06)	2049 (51.51)	< 0.001
	Yes	2,950 (35.00)	1,021 (22.94)	1929 (48.49)	
Food shortage	No	6,046 (71.74)	3,862 (86.79)	2,184 (54.90)	< 0.001
	Yes	2,382 (28.26)	588 (13.21)	1794 (45.10)	
Drug shortage	No	5,803 (68.85)	3,907 (87.80)	1896 (47.66)	< 0.001
	Yes	2,625 (31.15)	543 (12.20)	2082 (52.36)	
Degree of difficulty in daily household activities	No difficulty at all	2,602 (30.87)	1791 (40.25)	811 (20.39)	< 0.001
	Mild difficulties	3,803 (45.12)	2018 (45.35)	1785 (44.87)	
	Extreme difficulties	2023 (24.00)	641 (14.40)	1,382 (34.74)	
Perceived risks of infection	Low	1,560 (18.51)	964 (21.66)	596 (14.98)	< 0.001
	Medium	1738 (20.62)	914 (20.54)	824 (20.71)	
	High	5,130 (60.87)	2,572 (57.80)	2,558 (64.30)	

Chi-square test was used for balance checking.

drug shortage, perceived risk of infection, the degree of difficulty in daily household activities and infection risk level of living area), which were chosen based on the knowledge of the available literature related to the topics (11, 12, 22, 36). Finally, the adjusted multivariate logistic regression was introduced to analyze subgroup heterogeneity in sex, age, chronic disease, and actual infection risk level of the respondent's residence; the differences in self-care behavior outcomes between subgroups were tested using the Chow test (40). Statistical tests were considered significant if $p < 0.1$. The association between participant characteristics and study outcomes was quantified using standardized regression coefficients (β) and odds ratios (ORs) and their 95% CIs. All statistical analyses were performed using STATA statistical

software version 17.0 (StataCorp et al. Station 77,845, USA). $p < 0.05$ (two-sided) was considered statistically significant.

Results

Basic characteristics

Table 1 summarizes the characteristics of the sample of participants who completed "The 2020 China COVID-19 Survey" and illustrates whether participants' routine healthcare or medical consultation has been restricted due to COVID-19. A total of 8,428 participants were included; the average age was 32 years (SD 9.95 years, range 18–79 years), and 19.97% ($N = 1,683$) of them suffered from at least one chronic disease. In addition, 3,978 (47.2%) participants' routine medical care or medical consultations were restricted by COVID-19, and 9.31% ($N = 785$) of their family members got infected with COVID-19. Most of the differences in demographic and socioeconomic characteristics, health status, and COVID-19-related variables among the two RHSU subgroups were statistically significant.

Self-care behavior outcomes

The average total prevention behavior score for the groups in RHSU and control groups (Not RHSU) was 17.97 (SD = 3.54; 95% CI: 17.86–18.07; $p < 0.001$), 18.36 (SD = 3.34; 95% CI:18.26–18.45; $p < 0.001$), respectively (Figure 1). The mean Online medical consultation score was 1.51 (SD 1.34), used online consultation (52.63%), used video consultation (32.71), used telephone consultation service (44.23), used mail-order or personal delivery pharmacy (21.39%) and none used (36.22%). The data from Figure 1 showed that during the COVID-19 pandemic, 42.31% of participants engaged in weight control and 62.57% in physical activity. Compared to the control group, the RHSU group performed significantly better in weight control (proportion of responded "yes": 51.96% vs. 33.66%; $p < 0.001$), physical activity (proportion of responded "yes": 66.64% vs. 58.92%; $p < 0.001$) and online medical consultation (proportion of above-average score: 59.63% vs. 44.31%; $p < 0.001$).

The results of Multiple logistic regression analyses are shown in Table 2, we observed the RHSU was positively associated with prevention behavior (Coef = 0.16, 95% CI 0.003–0.321, $p = 0.045$), weight control (OR = 1.34, 95% CI 1.21–1.49, $p < 0.001$), physical activity (OR = 1.14,95% CI 1.03–1.27, $p < 0.001$), and online medical consultation (OR = 1.30, 95% CI 1.17–1.45, $p < 0.001$). From Table 2, we observed that participants aged 45 and over, female, married, non-chronic population, low-income groups, individuals who have not experienced COVID-19 infection (themself or family members), those who lived in low infection-risk areas, those whose daily household activities were not experienced difficulty due to COVID-19-related financial strain, and those with a perceived higher risk of infection were significantly more likely to report higher prevention behavior scores.

The average overall score for online medical consultation and prevention behavior was 1.51 (SD = 1.34, 95% CI:1.48–1.54) and 18.17 (SD = 3.44, 95% CI:18.10–18.24). In Table 2, multi-variable regression analyses indicated that urban residents, females, high-income groups, chronic disease population, those who experienced job loss due to the pandemic (participants or family members), those who lived in high infection-risk areas, those with a history of COVID-19 infection (participants or family members), those with better self-rated health, those who experienced food or drug

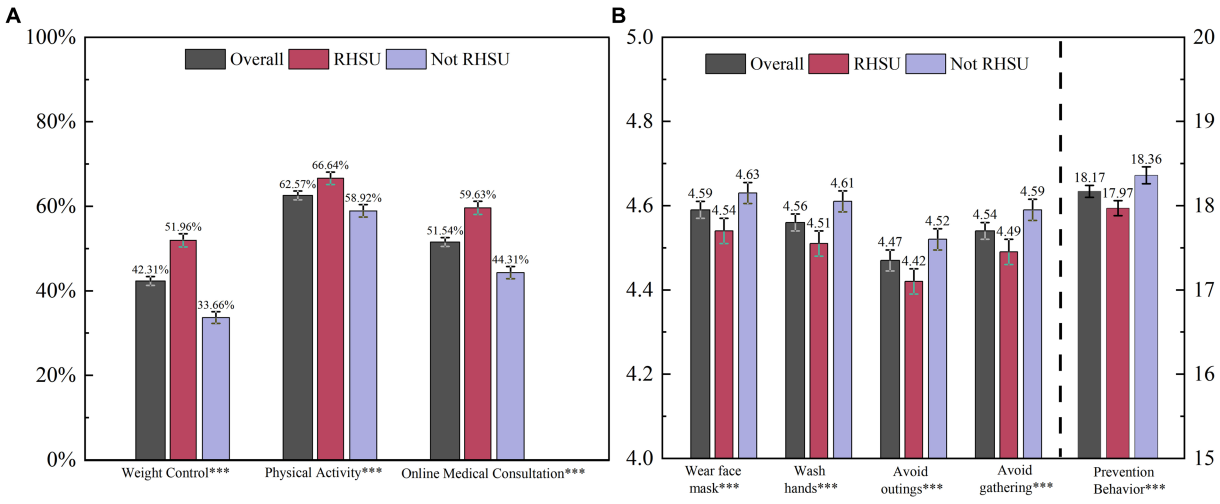

FIGURE 1
Self-care behavior outcomes between groups with or without restricted health service utilization among Chinese adults. **(A)** Shows the proportion of participants who engaged in weight control or physical activity and the proportion of above-average online medical consultation scores across the different groups. **(B)** Shows the mean of the total prevention behavior score and the score of each item. The total prevention behavior score ranges from 4 to 20, and the score of each item (i.e., wear face mask, wash hands, avoid outings, avoid gathering) ranges from 1 to 5. Higher scores mean better prevention behavior. The "Overall" represents the total sample, "Not RHSU" as the control group represents the participant's routine medical care or medical consultations have not been restricted due to COVID-19 and "RHSU" represents the participant's routine medical care or medical consultations have been restricted due to COVID-19. The error bars indicate a 95% confidence interval of the estimates. *** $p < 0.01$, ** $p < 0.05$, * $p < 0.1$.

TABLE 2 Associations of RHSU with each self-care behavior outcomes among Chinese adults.

Variables	Prevention behavior[a]		Online medical consultation[b]		Weight control[b]		Physical activity[b]	
	Coef (95%CI)	*p*-value	OR (95%CI)	*p*-value	OR (95%CI)	*p*-value	OR (95%CI)	*p*-value
Restricted Health Service Utilization (Ref. = No)	-		-		-		-	
Yes	**0.16 (0.00,0.32)**	**0.045**	**1.30 (1.17,1.45)**	**< 0.001**	**1.34 (1.21,1.49)**	**< 0.001**	**1.14 (1.03,1.27)**	**0.014**
Region (Ref. = City)	-		-		-		-	
Rural	−0.09 (−0.26,0.08)	0.284	1.12 (0.00,1.26)	0.052	**0.86 (0.77,0.97)**	**0.011**	0.98 (0.88,1.10)	0.760
Town	−0.17 (−0.38,0.04)	0.106	1.04 (0.91,1.20)	0.561	**0.78 (0.68,0.89)**	**< 0.001**	0.95 (0.83,1.09)	0.461
Actual infection risk level of living area (Ref. = Low)	-		-		-		-	
Moderate	**−0.25 (−0.41,-0.09)**	**0.002**	**1.16 (1.04,1.29)**	**0.007**	0.95 (0.86,1.06)	0.384	0.90 (0.81,1.00)	0.053
High	**−0.23 (−0.45,-0.02)**	**0.036**	**1.89 (1.62,2.21)**	**< 0.001**	**1.24 (1.07,1.43)**	**0.004**	1.16 (0.99,1.35)	0.061
Married status (Ref. = Unmarried/divorced/widowed)	-		-		-		-	
Married	**0.58 (0.43,0.73)**	**< 0.001**	**1.46 (1.31,1.62)**	**< 0.001**	0.94 (0.85,1.05)	0.265	**1.35 (1.22,1.49)**	**< 0.001**
Sex (Ref. = Female)	-		-		-		-	
Male	**−0.38 (−0.52,-0.24)**	**< 0.001**	**1.38 (1.25,1.52)**	**< 0.001**	**0.86 (0.78,0.94)**	**< 0.001**	**1.32 (1.20,1.45)**	**< 0.001**
Age (years) (Ref. = 18–44)	-		-		-		-	
≧45	**0.32 (0.09,0.54)**	**0.005**	**0.35 (0.30,0.41)**	**< 0.001**	0.96 (0.83,1.12)	0.633	**0.84 (0.73,0.97)**	**0.020**
Bachelor degree (Ref. = No)	-		-		-		-	
Yes	0.12 (−0.03,0.26)	0.128	1.09 (0.99,1.21)	0.092	0.98 (0.88,1.08)	0.672	1.05 (0.95,1.17)	0.322
Household income Level (Ref. = Low)	-		-		-		-	
Medium	−0.16 (−0.34,0.02)	0.076	**1.40 (1.24,1.58)**	**< 0.001**	0.98 (0.87,1.10)	0.714	**1.39 (1.23,1.56)**	**< 0.001**
High	**−0.31 (−0.47,-0.14)**	**< 0.001**	**1.52 (1.36,1.71)**	**< 0.001**	**1.23 (1.10,1.37)**	**< 0.001**	**1.25 (1.12,1.40)**	**< 0.001**
Self- rated health status (Ref. = Fair or poor)	-		-		-		-	
Good	0.13 (−0.32,0.58)	0.576	1.02 (0.75,1.39)	0.915	1.31 (0.96,1.79)	0.086	**1.69 (1.25,2.29)**	**< 0.001**
Very good	0.30 (−0.12,0.73)	0.163	**1.86 (1.39,2.49)**	**< 0.001**	**1.83 (1.36,2.45)**	**< 0.001**	**4.17 (3.12,5.57)**	**< 0.001**
Chronic disease groups (Ref. = No)	-		-		-		-	
Yes	**−1.01 (−1.20,-0.83)**	**< 0.001**	**1.54 (1.35,1.76)**	**< 0.001**	**1.17 (1.04,1.33)**	**0.009**	0.93 (0.82,1.05)	0.247

(Continued)

TABLE 2 (Continued)

Variables	Prevention behavior[a]		Online medical consultation[b]		Weight control[b]		Physical activity[b]	
	Coef (95%CI)	*p*-value	OR (95%CI)	*p*-value	OR (95%CI)	*p*-value	OR (95%CI)	*p*-value
COVID-19 infection (participants or family member) (Ref. = No)	-		-		-		-	
Yes	**−2.01 (−2.28,-1.74)**	**< 0.001**	1.03 (0.84,1.26)	0.786	**2.28 (1.88,2.76)**	**< 0.001**	**1.70 (1.38,2.09)**	**< 0.001**
Lost job due to COVID-19 (Ref. = No)	-		-		-		-	
Yes	0.10 (−0.07,0.27)	0.243	1.05 (0.93,1.18)	0.435	**1.34 (1.20,1.50)**	**< 0.001**	1.12 (0.99,1.26)	0.056
Food shortage (Ref. = No)	-		-		-		-	
Yes	−0.17 (−0.36,0.01)	0.070	**1.19 (1.04,1.35)**	**0.008**	**1.46 (1.30,1.65)**	**< 0.001**	**1.26 (1.11,1.43)**	**< 0.001**
Drug shortage (Ref. = No)	-		-		-		-	
Yes	0.00 (−0.18,0.18)	0.995	**1.56 (1.37,1.76)**	**< 0.001**	**1.43 (1.27,1.61)**	**< 0.001**	**1.37 (1.21,1.55)**	**< 0.001**
Degree of difficulty in daily household activities (Ref = No difficulty at all)	-		-		-		-	
Mild difficulties	**−0.31 (−0.48,-0.13)**	**< 0.001**	**1.94 (1.73,2.18)**	**< 0.001**	1.06 (0.94,1.19)	0.338	0.93 (0.83,1.04)	0.180
Extreme difficulties	**−0.76 (−1.00,-0.53)**	**< 0.001**	**2.38 (2.03,2.80)**	**< 0.001**	**1.31 (1.12,1.53)**	**< 0.001**	1.00 (0.85,1.18)	0.971
Perceived risks of infection (Ref. = Low)	-		-		-		-	
Medium	**0.35 (0.13,0.57)**	**< 0.001**	**0.60 (0.52,0.71)**	**< 0.001**	0.954 (0.81,1.10)	0.454	1.00 (0.86,1.16)	0.994
High	**1.25 (1.06,1.44)**	**< 0.001**	**0.38 (0.33,0.43)**	**< 0.001**	0.88 (0.78,1.00)	0.053	**0.85 (0.74,0.96)**	**0.011**

Coef., Coefficient; OR, Odds Ratio; CI, confidence interval. [a]denotes continuous dependent variable, [b]denotes binary dependent variable, statistically significant results are in bold ($p<0.05$).

shortages, and those whose daily household activities experienced extreme difficulties due to COVID-19-related financial strains, were significantly more likely to engage in weight control. Meanwhile, the young group (18–44 years), male, married, medium and high-income groups, those with better self-rated health, those with a history of COVID-19 infection (participants or family members), and those who experienced food or drug shortage were significantly more likely to engage in physical exercise. Also, the results showed that the young group (18–44 years), male, married, medium and high-income groups, chronic disease population, those with very good self-rated health, those who lived in higher infection-risk areas, those experienced food or drug shortages, those whose daily household activities were experienced mild or extremely difficulties due to COVID-19-related financial strain, and those who perceived high risk of infection were significantly more likely to report above-average online medical consultation score.

Subgroup analysis

Based on the relevant literature, we generated multiple logistic regression models that included all variables to conduct subgroup analyses among sex, age, chronic disease, and actual infection risk level of the respondent's residence (41). According to the subgroup analysis shown in Figure 2, we observed significant positive associations between RHSU and four indicators of self-care behavior (weight control, physical activity, prevention behavior, and online medical consultation) in all populations. By conducting a Chow test on the coefficients of the subgroup regression, we found that male groups (Male, OR = 1.33, CI 1.06–1.1.67, *Chow text* $p = 0.016$) and participants living in high-risk areas were more likely to engage in physical activity (high infection-risk level, OR = 1.51, CI 1.03–1.27, *Chow text* $p = 0.024$) when RHSU occurred. Similarly, when both groups experienced RHSU, participants living in higher infection-risk

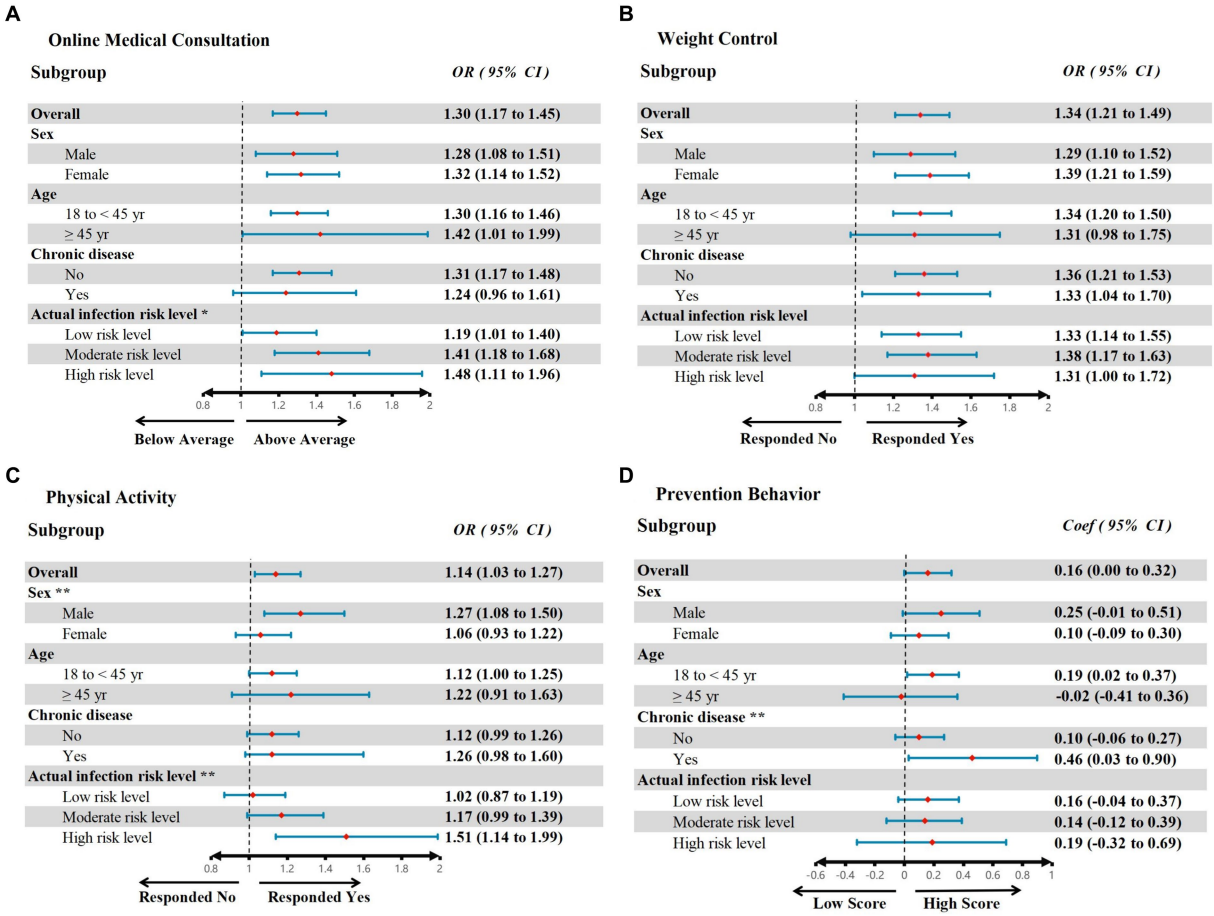

FIGURE 2
Associations of Restricted Health Service Utilization and Self-Care Behavior outcomes among subgroups. The red dots represent the observed mean value, and the blue lines represent the odds ratio **(A–C)** or coefficient **(D)** in the adjusted model; the CHOW test tested all outcomes of the subgroup analyses, ** Chow text $p < 0.05$, * Chow text $p < 0.1$. Adjust model control variables, i.e., age, sex, region, married status, actual infection risk level of living area, educational level, household income level, history of COVID-19 infection (participants or family member), chronic disease, self-rated health, food and drug shortage, lost job due to COVID-19, daily activities affected by COVID-19 related financial strain and perceived risk of infection. The Appendix File 2 shows details of multiple regression models for sex, age, chronic disease, and infection risk level of the living area subgroup.

areas were more likely to report above-average online medical consultation scores (high infection-risk level, OR = 1.48, CI 1.03–1.27, *Chow text* $p = 0.078$; moderate infection-risk level, OR = 1.41, CI 1.18–1.68, *Chow text* $p = 0.098$), In addition, the chronic disease group (Coef = 0.46, CI 0.03–0.90, *Chow text* $p = 0.032$) was more likely to report higher prevention behavior scores. Conversely, the relation between RHSU and weight control was not statistically different in subgroup analyses based on sex, age, chronic disease, and actual infection risk level of the respondent's residence.

Discussion

Since March 2020, the World Health Organization declared COVID-19 a global pandemic, and governments around the world imposed restrictions on the use of hospitals and outpatient services, eliminated all elective, routine and non-emergency patient procedures, implemented stricter physical distancing measures and transitioned to remote care to reallocate resources to the urgent care of patients with COVID-19 (42), which substantially disrupted individuals' routine healthcare utilization. Globally, the Chinese government's endeavor to contain the spread of the COVID-19 pandemic has been widely praised, which may have contributed to a more severe influence on individuals' routine healthcare utilization during the early stages of the outbreak compared to other countries (9). To the best of our knowledge, this is the first study to explore the impact of health service utilization on self-care behavior among Chinese adult populations during the early COVID-19 pandemic based on a large sample covering 31 provinces in China. Furthermore, our study used two dimensions with four indicators to measure self-care behavior, explore the relationship between RUSH and self-care behavior, and examine vulnerable populations and subgroup differences through multilevel regression and subgroup analyses. Our findings from the COVID-19 pandemic may further elucidate the future policy development of self-care behavior in China, provide appropriate interventions to mitigate the health risks caused by RHSU and contribute to alleviating the burden on the public health system and its long-term benefits.

Firstly, the multivariate results supported much of what we anticipated earlier: the presence of RHSU was positively associated

with all four kinds of self-care behavior in this study, and these associations remained significant even after adjusting for individual, environmental, and risk-perceptive control variables. Similar to the results of studies before (like medical services supply decline) and after the outbreak of the COVID-19 pandemic, these researchers also observed the association between social inequalities, pandemic-related changes, and individual health behavior (41–43). For instance, when individuals have potential health risks (including reduced accessibility to healthcare or deterioration of health status), this may stimulate the self-care activities and behaviors that were aimed at preventing or reducing health risks and optimizing health and quality of life (22, 44). Also, evidence from Anderson's health behavior model likewise supports a relationship between the use of preventive health services and self-care ability in daily life (46). Regarding autonomous behavior, previous studies claim that there is an association between decreased healthcare utilization and increased leisure-time physical activity; this association remains after adjustment for socio-economic confounders (17), which is consistent with our findings. Research with adults during the COVID-19 pandemic suggests that self-isolation at home due to lockdown is associated with a lower level of physical activity and modifications in eating behavior (47, 48). Also, a classic literature review elucidated that individual age, health literacy, and self-rated health could indirectly influence the association between healthcare utilization and physical activity through mediation analyses (49). Based on the discussion above, we predict that the relationship between self-care behavior and RHSU will persist even after the COVID-19 pandemic subsides.

Secondly, even if there is a willingness to practice positive self-care behavior, some vulnerable people still perform worse in familiarizing self-care activities and building self-care behavior due to physical and psychological factors (26), which if ignored could make it difficult to implement the entire strategy of reducing the burden on public health through self-care behavior. For example, previous studies indicated that 30–49-year-old adults, those with a higher level of education, and those who were employed and had a high income were more knowledgeable and better able to take appropriate measures to prevent the spread of COVID-19 (35, 50). However, our study observed that participants in the survey had generally high scores on prevention behaviors related to COVID-19, and these differences between populations were not apparent compared to other studies, which may be attributed to the Chinese government's extensive publicity and appropriate supervision of interrupting the spread of the disease in the early stages of the COVID-19 pandemic (51).

Similarly, our study indicated significant correlations in online medical consultation across age, gender, income, married status, health status, and infection risk perception, those findings align with previous studies (27, 52). It is not surprising that social distance, isolation, and hospital restrictions forced chronic patients to re-organize their routine care through temporary in-person visits during the pandemic, this led to a widespread and significant increase in telemedicine utilization (3). Also, patients with chronic disease may avoid in-person visits to hospitals, clinics, and emergency departments for fear of exposure to potential COVID-19-infected patients (3, 53), thereby preferring to implement telemedicine or seek online medical advice. However, our results showed that middle-aged and older adults showed relatively weak performance in medical consultation compared to younger adults. In fact, despite research examining age differences in self-care during the COVID-19 pandemic has been inconclusive, previous studies on the digital divide demonstrated that people aged above 65 years are less likely than the younger generation to have had the chance to familiarize themselves with ICT either at school or at work, combined with the cognitive, motor, digital gap and sensory decline associated with aging, and older adults face more barriers to and challenges in using online technology for health than their younger counterparts (54). The present studies claimed that this digital divide created by digital technologies might widen social inequalities by alienating disadvantaged groups that do not have access to digital resources (55, 56). Thus, we recommend that particular attention be paid to the older adult when future discussions on critical issues surrounding the promotion of online medical consultations include quality of care (57), communication and language barriers (58), and patient satisfaction (43).

Thirdly, our study also extended the existing literature by the correlations between restricted health service utilization and self-care behavior, which were quantified and compared by subgroup analysis. Based on this, we found that external and intrinsic risk factors had a significant effect on some type of self-care behavior. For example, residents living in the higher infection-risk areas were more likely to report above-average online medical consultation scores or engage in physical activities when they experienced RHSU. The strength of this association varies significantly with the infection risk level of the living area. These findings are also consistent with the argument for behavioral change theory since high infection-risk living environments created additional barriers (e.g., extra healthcare costs, and psychological stress) to illness prevention and health maintenance. Therefore, except for inequities in health service utilization, the risk perception (41) and actual infection risk related to the pandemic (44) may be critical indicators of health behavior choices. A possible explanation from psychology is that the higher the risk an individual perceives, the more motivated they might be to engage in protective behaviors (53). Despite this, both external and intrinsic risk factors need to be considered for personal characteristics, previous research highlighted that not everyone responds to health risks similarly and that risk perception alone does not explain health behavior (59). For instance, the self-care behavior of chronic disease patients is associated with a high perceived susceptibility to disorders; they may tend to avoid in-person visits to hospitals, clinics, and emergency departments for fear of exposure to potential COVID-19 (43). Likewise, older adults are more vulnerable to COVID-19 infection, have a worse prognosis after infection, and have a higher risk of getting one or more non-communicable diseases, so that they may experience heightened levels of instilled fear of COVID-19 exposure during in-person medical services (3). In general, caution must be exercised in the following discussion of these findings.

In contrast, no significant associations were found in the model of weight control among subgroups analysis, which means that the strength of the association between RHSU and weight control was not affected by age, chronic diseases, and actual infection risk level of the respondent's residence. Still, we found several reasons that may confound our findings by searching the literature. A study from *Obesity* has suggested that social closure measures may have a wide-ranging effect, making it more difficult for many people to adopt weight gain protective behaviors (60). It was widespread for people to experience barriers to diet and healthy eating during the COVID-19 pandemic lockdown (43, 47) (e.g., food shortage, lacking motivation and control around food). Also, some mental health issues, such as

anxiety, stress, and poor mood, were found to be risk factors for obesity during the pandemic (61). Collectively, the factors mentioned above may make it more difficult to distinguish differences in weight control behaviors among populations.

At last, given that individuals who experienced restricted health service utilization were more actively engaged in self-care behavior, our study examines the interaction of health service inequalities and potential risk factors on self-care behavior during the pandemic. It highlights the necessity of promoting self-care behavior when in response to health service restrictions, especially for vulnerable individuals (62). Meanwhile, in view of the various benefits of self-care behavior, we need more comprehensive policies to encourage individuals to develop self-care behavior. Both primary healthcare institutions and community service organizations should strengthen the strategic support related to self-care promotion and implementation for the public regarding personal health literacy development, public healthcare services, online healthcare services, continuing medical education, etc.

From the perspective of health care system reform, addressing current barriers around self-care in terms of applicability, developmental disequilibrium, standardization, supervision, service coverage, and digital divide caused by technological advances will help more people maintain their health through self-care activities effectively; it not only effectively responds the issue of restricted health service utilization, but also contributes to alleviate the burden on the public health system and its long-term benefit (63). Indubitably, there is a pressing need to strengthen the development of self-care systems in China.

Our study includes several fundamental limitations that must be acknowledged and addressed in future studies. Given the cross-sectional nature of this study, one of the main limitations was that there was no baseline response rate before the pandemic and no available data on participants' previous self-care behavior; neither can make definitive statements about causality in regression analyses. Second, this study assessed outcome variables by employing a few single-item scales; also, only three dimensions were collected to describe the performance of self-care behavior. Therefore, future research should determine variables more comprehensively by using multiple scales to provide more conclusive evidence on the predictive validity of self-care behavior. Third, participants were recruited using a snowball sampling method through social media; the advantage of this method is that a large number of samples can be collected quickly. However, many participants were well-educated and below 40 years old, leading to a particular bias in the results, which made it difficult to identify more subgroups with significant differences. Fourth, the subgroup analyses in this study did not use more demographic categories, such as region, economic status, and education level; these subgroup differences require deeper exploration in the future.

However, this study has some innovative findings. First, our findings extend the existing literature by exploring the impact of restricted health service utilization on self-care behavior in a large sample covering all provinces in mainland China. Based on the RAA theory, our study may find some motivations and factors for self-care behavior change; these may provide some references for promoting the effectiveness of self-care behavior to reduce the burden on the health service system. Second, in contrast to previous studies mainly focusing on the impact of inequalities in health service accessibility or quality on self-care behavior in the context of regional economic disparities, our study contributes to the existing knowledge base by investigating the relationship between restricted health service utilization and self-care behavior during a large scale infectious disease crisis. Third, we also take into account differences in risk perception when exploring the impact of inequalities in health service utilization on the outcome variables, so we tried to describe and compare the association between the RHSU and self-care behavior among the sex, age, chronic disease and high, middle or low risk of infections living area subgroups, it is effective in terms of filling the gap in the relevant literature.

Conclusion

The COVID-19 pandemic dramatically impacted health service utilization across China (9), and this study sheds light on the restricted health service utilization may predict more positive self-care behavior during the pandemic and the differential presentation of this association between subgroups of sex, age, chronic disease, and actual infection risk level of the residence area. Based on our results and current research findings, we believe that the correlation between restricted health service utilization and self-care behavior will persist as the COVID-19 pandemic subsides. Thus, we need further research into the mechanisms of self-care behavior, as well as continuing to address self-care knowledge gaps and improve outcomes. Given the many existing challenges to our vision, it is necessary to drive the development of policies related to self-care behavior to raise self-care as a vital element in general health and healthcare, which not only effectively responds the adverse health outcomes from restricted health service utilization and future public health crisis, but also contributes to alleviating the burden on the healthcare system and its long-term benefits.

Data availability statement

The data analyzed in this study is subject to the following licenses/restrictions: the datasets presented in this article are not readily available because ethics restrictions. Requests to access these datasets should be directed to youfawang@gmail.com.

Ethics statement

The studies involving humans were approved by the Institutional Review Committees of the Xi'an Jiaotong University, China (approval number 2020-1172). 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

ZW: Conceptualization, Data curation, Formal analysis, Resources, Software, Writing – original draft, Writing – review & editing. ZZ: Data curation, Formal analysis, Funding acquisition, Writing – review & editing, Supervision. GL: Investigation, Methodology, Project administration, Writing – original draft. JL:

Resources, Writing – review & editing, Formal analysis, Investigation. XZ: Methodology, Writing – review & editing, Resources, Validation. XF: Data curation, Investigation, Writing – review & editing, Methodology. SL: Conceptualization, Data curation, Investigation, Writing – review & editing. YW: Conceptualization, Data curation, Investigation, 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.2024.1398271/full#supplementary-material

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Sociodemographic disparities in postnatal care coverage at comprehensive health centers in Hamedan City

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Background: Postnatal care (PNC) is a crucial component of continuous healthcare and can be influenced by sociodemographic factors. This study aimed to examine the sociodemographic disparities in PNC coverage in Hamedan City.

Methods: In this cross-sectional study, we utilized existing data recorded in the Health Integrated System of Hamedan City, located in Iran, from 2020 to 2021. The study population consisted of 853 women who were over 15 years old and had given birth within the past 42 days. The Health Equity Assessment Toolkit (HEAT) software was used to evaluate the socioeconomic inequalities in PNC coverage.

Results: Overall, 531 (62.3%) of the women received three postnatal visits. The absolute concentration index (ACI) indicates that women aged 20–35 years, illiterate women, housewives, insured individuals, and urban residents experience a higher magnitude of inequality in PNC coverage. The negative values of the ACI suggest that the health index is concentrated among disadvantaged groups, with educational level inequalities being more pronounced than those related to age.

Conclusion: Postnatal care coverage among mothers was relatively adequate; however, sociodemographic inequalities existed in the utilization of PNC services. It is recommended that policymakers make efforts to increase access to PNC services for mothers from low socio-economic groups.

KEYWORDS

postnatal care coverage, midwifery services, women health, Iran, disparities - definition and paradigm

Introduction

Most maternal deaths occur during the postnatal period, and an estimated 2.8 million babies die within the first month of life (1). Postnatal care (PNC) services are crucial for improving outcomes for mothers and infants in low- and middle-income countries (2). Inadequate PNC can expose mothers to risks such as postpartum bleeding, eclampsia, puerperal

infection, thromboembolic disease, breastfeeding problems, and psychological issues such as depression (3). In some countries, PNC coverage is relatively poorer compared to other maternal and child care services (4). The World Health Organization (WHO) recommends that mothers and their newborns receive postnatal care within 24 h of birth, on the third day, during the second week, and at 6 weeks postpartum (5).

In Iran, postpartum care is typically provided at governmental and nongovernmental facilities, with all services delivered outside the home. Governmental postnatal care services are provided free of charge and include the prevention, early detection, and treatment of complications and diseases, as well as counseling on breastfeeding, birth spacing, immunization, and maternal nutrition (6). Improving social conditions and facilitating access to health services and education can play a valuable role in ensuring the health of mothers and children, who are among the most vulnerable groups.

Considering the necessity of PNC as an essential strategy to save the lives of mothers and newborns, it is crucial to identify the factors that prevent mothers and infants from benefiting from postnatal care (6). Health inequality, defined as the disproportionate concentration of people with specific health behaviors in certain demographic subgroups, remains a significant challenge for health systems, especially in low-income countries (7). For instance, mothers with lower levels of education and income are much less likely to initiate and continue breastfeeding than those from higher socioeconomic classes (8).

Global research has identified various factors influencing the use of PNC services, with varied outcomes, including maternal age, education level, occupation, place and method of delivery, number of pregnancies, and awareness of PNC services (9, 10). Economic and social determinants can affect the receipt of these essential services or lead to adverse health consequences during the postnatal period. Individuals with lower socioeconomic status are less likely to receive routine healthcare or preventive health advice and have a higher incidence of adverse health outcomes (11, 12).

The results of the FiNaL Study showed that women belonging to low socioeconomic levels face significant deprivations in terms of access to formal and informal breastfeeding support and even access to information (13). A study conducted in Nigeria classified the determinants of PNC services into family and community levels, which include education level, financial status, urban or rural residence, religion, source of information, mother's age, and previous experience with health services (14). Another study on determinants affecting maternal health outcomes in Ghana revealed that rural residents are less likely than urban residents to undergo PNC (15).

It is important to note that the variables affecting PNC service use differ based on socio-cultural factors within a particular community. These differences may be attributed to factors such as access to health facilities, intentional government interventions, geographic location, and cultural practices. Identifying sociodemographic factors associated with PNC utilization can help health planners design and implement evidence-based interventions to strengthen the health system and improve access to and use of PNC services. This study aimed to investigate the social and economic inequalities in postnatal care coverage among mothers in Hamedan City.

Abbreviations: PNC, Postnatal care; HEAT, Health Equity Assessment Toolkit; R, Ratio; ACI, Absolute Concentration Index; DHS, Department of Homeland Security; WHO, World Health Organization.

Methods

Study design and setting

In this cross-sectional study, existing data from the Health Integrated System of Hamedan City, located in Iran, for the years 2020–2021 were analyzed to assess socioeconomic inequalities in postnatal care (PNC) coverage. In Hamedan city, comprehensive health coverage is provided to all residents, both urban and rural, through health centers. All healthcare information, including PNC, is electronically recorded in the integrated health system. According to national guidelines, postpartum care includes three visits on days 1–3, 10–15, and 42–60 after delivery.

Participants

The study population comprised women over 15 years old who had given birth within the past 42 days. A total of 853 women met these criteria and their data were included in the analysis.

Sampling method

In Hamedan City, there are 17 comprehensive urban and 13 comprehensive rural health centers. For this study, a simple random sampling method was used to select 8 out of the 17 comprehensive urban health centers and 5 out of the 13 comprehensive rural health centers. Subsequently, all eligible participants within the selected centers were enrolled using the census sampling method.

Data collection tools

Demographic and obstetrics checklist

The checklist included the following variables: age, occupation, education level, place of residence, insurance status, delivery method, experience of preterm delivery, and exclusive breastfeeding.

Postnatal coverage

The primary outcome of the present study is the number of postnatal visits within 42 days after delivery, which ranges from 0 to 3 visits according to national guidelines.

Data analysis

Data analysis was performed using SPSS software version 16. Descriptive statistics were used to summarize the data. The relationship between qualitative variables was assessed using the Chi-square test, and a logistic regression model was employed to identify predictive variables associated with receiving postnatal care (PNC) at a 95% confidence level.

To assess inequalities in PNC coverage across socio-economic subgroups, the Health Equity Assessment Toolkit (HEAT) software version 4.0 (Beta) was utilized. Inequality was evaluated using the ACI (absolute concentration index) and R indices. The R ratio is a simple measure indicating relative inequality between two subgroups, with values greater than 1 indicating higher inequality (16). The ACI is a weighted measure that assesses inequality based on a natural ordering

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scale, where positive values indicate concentration among advantaged groups and negative values indicate concentration among disadvantaged groups. A higher absolute ACI indicates greater inequality (17).

Results

Demographic characteristics and their related factors

The results of data analysis indicated that the highest frequency distribution was observed among individuals aged 20–35 years (74.7%), unemployed individuals (91.3%), those who were illiterate (43.3%), urban residents (82.2%), and those with insurance coverage (95.3%).

Analysis of postnatal care (PNC) coverage based on demographic characteristics revealed that the highest frequency distribution of receiving at least one or more PNC visits was among individuals aged 20–35 years (74.4%), unemployed individuals (91.9%), those who were illiterate (44%), urban residents (82.1%), and those with insurance coverage (95.2%).

Furthermore, comparing PNC coverage according to employment status showed statistically significant differences ($p = 0.014$) (Table 1).

TABLE 1 Socio-economic characteristic of women in term of postnatal visits ($N = 853$).

Characteristics *N* (%)		Postnatal visits, *n* (%)		*p*-value
		0	1–3	
Age (y)				0.38
<20	67 (7.9)	1 (2.4)	66 (8.1)	
20–35	637 (74.7)	34 (81)	603 (74.4)	
>35	149 (17.5)	7 (16.7)	142 (17.5)	
Job				0.01*
No	779 (91.3)	34 (81%)	745 (91.9)	
Yes	74 (8.7)	8 (19%)	66 (8.1)	
Education				0.12
Illiterate/ Elementary	369 (43.3)	12 (28.6%)	357 (44)	
Secondary	244 (28.6)	14 (33.3%)	230 (28.4)	
Higher	240 (28.1)	16 (38.1%)	224 (27.6)	
Location Residence				0.84
Rural	152 (17.8)	7 (16.7%)	145 (17.9)	
Urban	701 (82.2)	35 (83.3%)	666 (82.1)	
Insurance				0.46
No	40 (4.7)	1 (2.4%)	39 (4.8)	
Yes	813 (95.3)	41 (97.6%)	772 (95.2)	

Postnatal coverage and its related factors

More than 62% (531) of mothers received all three postnatal care visits. However, less than 5% (42) of women did not receive any postnatal care. The rate of exclusive breastfeeding up to 42 days after childbirth was 94.4% (805) (Table 2).

The prevalence of cesarean section was 53.2% (454), preterm labor was 7.6% (65), and unwanted pregnancy was 19.1% (690). Cesarean section was significantly associated with the number of postnatal care (PNC) visits, with a higher percentage of individuals not receiving any PNC at all following cesarean section compared to normal vaginal delivery (Table 3).

A logistic regression model was employed to identify predictive variables associated with postnatal care (PNC). The results indicated that, after controlling for variables such as age, education, insurance, and residence of mothers, occupation showed a significant relationship with the frequency of PNC visits. Specifically, the odds of receiving care were 5.5 times higher among non-employed mothers compared to employed mothers ($p = 0.018$) (Table 4).

TABLE 2 The frequency of Postpartum Visits and Breast Milk Feeding (BMF) coverage.

Characteristics	*N* (%)
Postpartum visits	
0	42 (4.9)
1	101 (11.8)
2	179 (21.0)
3	531 (62.3)
Breast Milk Feeding (BMF)	
No	48 (5.6)
Yes	805 (94.4)

Socio-economic inequalities and PNC coverage

The results from Table 5 indicate that the coverage of at least one postnatal care visit was 95%. The absolute concentration index (ACI) highlights that the age group 20–35 years exhibits a higher magnitude of inequality in postnatal care (PNC) coverage compared to other age groups. Specifically, PNC coverage was more equitable in the age groups under 20 and over 35 years old.

Regarding other demographic characteristics, inequalities in PNC coverage were more pronounced among the illiterate, housewives, insured individuals, and urban residents compared to other subgroups.

Furthermore, the R indices (R) revealed that the level of inequality was greater for the "insurance" variable ($R > 1$) and lower for variables such as age, education, occupation, and place of residence ($R < 1$). This suggests that inequality in PNC coverage is more pronounced based on insurance status compared to other demographic factors.

Negative values of the ACI indicate that the concentration of the health index is among disadvantaged populations, with educational inequalities being more prominent than age-related disparities (Table 5).

Discussion

The results of the present study revealed that postnatal care (PNC) coverage in Hamedan, a city in Iran, was moderately high, with more than 62% of postpartum mothers receiving PNC, aligning with World Health Organization recommendations. Evidence suggests that widespread PNC availability could prevent between 10 and 27% of infant deaths (18). In this study, 37.7% of mothers received care 1–2 times, while approximately 5% did not receive any PNC at all. Limited

TABLE 4 The logistic regression model of the postnasal visits and demographics characteristics.

Backward model		B	S.E.	Wald	*p*-value[a]	Exp (B)	95% CI	
							Lower	Upper
Postnatal care coverage	Job (yes)	ref						
	Job (no)	0.977	0.413	5.583	0.01*	2.656	1.181	5.972

[a]Adjusted to age, education, location, and insurance variables.

TABLE 3 The relationship between postnatal visits with obstetric characteristics of women (*N* = 853).

Characteristics *N* (%)		Postnatal visits, *n* (%)		*p*-value
		0	1–3	
Mode of delivery				<0.001*
CS	454 (53.2)	32 (7)	422 (93)	
NVD	399 (46.8)	10 (2.5)	389 (97.5)	
Intention to get pregnant				0.10
Unplanned	690 (80.9)	38 (5.5)	652 (94.5)	
Planned	163 (19.1)	4 (2.5)	159 (97.5)	
Preterm labor				0.18
No	788 (92.4)	41 (5.2)	747 (94.8)	
Yes	65 (7.6)	1 (1.5)	64 (98.5)	

CS, Cesarean section; NVD, Normal vaginal delivery.

PNC coverage is evident, particularly among economically disadvantaged families. For instance, in the Democratic Republic of the Congo, only 35% of women receive PNC (19), while in Nepal, PNC utilization stands at around 22% (20). In contrast, a study in Sri Lanka reported 76.9% coverage of recommended PNC visits (21).

In Iran, studies on PNC coverage are scarce, with data typically managed by the Ministry of Health. However, a longitudinal study in Tehran found high rates of prenatal care utilization (95%) and recommended visits (99%) among pregnant women (22).

Notably, a significant proportion of deliveries in the present study were by cesarean section (53.2%), and these mothers were less likely to receive PNC compared to those with vaginal deliveries. Similar findings on socio-economic inequality in PNC utilization after cesarean sections were reported in other studies (23), although some research in Ethiopia suggests cesarean delivery may increase PNC utilization due to perceived higher risks (24).

Iran has one of the highest cesarean section rates globally (25). Most cesarean sections in this study were likely emergency procedures, underscoring the critical need for post-cesarean care and follow-up due to higher complication risks compared to vaginal deliveries (26). Financial and cultural barriers may hinder cesarean-section mothers from accessing PNC, as seen in studies from Pakistan highlighting transportation and healthcare costs as deterrents (27).

Regarding demographics, PNC coverage was better among women under 20 and over 35 years old compared to those aged 20–35 years. In Iran, younger mothers (<20 years) receive heightened attention due to perceived higher pregnancy risks, while older mothers (>35 years) benefit from greater healthcare awareness and support. Similar findings on increased PNC utilization with advancing maternal age have been reported elsewhere (28, 29).

Education also significantly influenced PNC coverage, with lower rates among illiterate or minimally educated women compared to others (24–29). Education enhances health awareness and promotes health-seeking behavior, influencing service utilization (30).

Non-employed mothers in the study were 5.5 times more likely to receive PNC compared to employed counterparts, contrary to some expectations about economic independence and service access (29, 31). Rural residents also showed higher PNC coverage than urban counterparts, differing from findings in some studies (32, 33), but consistent with others (20, 34). Urban areas may offer better access to private healthcare facilities and health promotion programs, influencing PNC utilization (21).

Interestingly, while 95.3% of mothers in this study had health insurance, uninsured individuals had better PNC coverage in comprehensive health centers. The availability of health insurance allows mothers to choose private facilities or gynecologists' offices, potentially reducing visits to public health centers. Efforts to improve PNC service quality in public centers could enhance overall access and utilization, regardless of socio-economic status.

Strengths of study

The study benefits significantly from using data from the Health Integrated System, which likely offers a large sample size. This enables a comprehensive analysis of postnatal care (PNC) utilization and inequalities. The data within this system are objective and reliable, systematically collected as part of the healthcare system. This enhances the credibility of the study findings and reduces biases associated with self-reported data or survey responses. As a result, the study provides more accurate and representative insights into disparities in postnatal care. These findings can have profound implications for healthcare policies and interventions aimed at reducing inequality in PNC, thereby contributing to improved maternal and child health outcomes.

Limitations of study

This study has several limitations that should be considered. Firstly, it is unable to examine disparities in postnatal care (PNC) provided at private facilities, focusing solely on public health centers. Secondly, as a cross-sectional analysis, it can establish associations between variables but cannot determine causation. Additionally, using secondary data introduces limitations such as potential exclusion of important factors like family support, proximity to healthcare facilities, and availability of

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TABLE 5 The percentage of affected population in terms of subgroups and inequality index.

Postnatal care coverage	Variable	Percentage of affected population	Risk Q1/Q5	Estimate of coverage	Ci 95% lower	Ci 95% upper	Aci	*R*	Setting average
	Age, y		0.45				−1.15	0.86	95
	<20	7.9		92.5	86	99			
	20–35	74.7		83.0	80.1	85.9			
	>35	17.5		79.8	73.3	86.3			
	Education		1.54				−3.87	0.80	
	Illiterate/ elementary	43.3		89.7	86.5	92.8			
	Secondary	28.6		84.8	80.3	89.3			
	Higher	28.1		71.6	65.9	77.4			
	Insurance		–				–	1.01	
	No	4.7		82.5	70.1	94.8			
	Yes	95.3		83.2	80.7	85.8			
	Location residence		–				–	0.89	
	Rural	17.8		91.4	86.9	95.9			
	Urban	82.2		81.4	78.5	84.3			
	Job		–				–	0.75	
	No	91.3		85.1	82.6	87.6			
	Yes	8.7		63.5	52.2	74.7			

ACI, Absolute concentration index; R, Ratio*.

healthcare providers. Maternal factors like residence patterns can also impact PNC access; for example, many Iranian mothers, particularly first-time mothers, often return to their parents' homes postpartum, seeking care from family members, especially their mothers.

Furthermore, the study's scope is limited to Hamedan City, a specific geographic area in Iran, which may restrict the generalizability of findings to other regions. To gain a comprehensive understanding of maternal and child health disparities, ongoing monitoring and evaluation of health indicators and related inequities are crucial. Future research that considers geographical and socio-economic factors could provide valuable insights into variations in postnatal care across diverse social and cultural contexts.

Conclusion

Our analysis revealed significant disparities in postnatal care (PNC) utilization, particularly among specific demographic groups such as women aged 20–35, those with low education levels, housewives, insured individuals, and urban residents. By elucidating the intricate relationship between socioeconomic status and PNC coverage, our study offers valuable insights into global health inequalities. The negative values of the absolute concentration index (ACI) underscore the concentration of inadequate PNC coverage among marginalized individuals, emphasizing the urgent need for targeted interventions at a systemic level.

As we strive for universal health coverage and work toward achieving the Sustainable Development Goals, the findings of this research hold relevance Hamedan City, extending to regions worldwide grappling with similar challenges in ensuring equitable maternal healthcare. Addressing socioeconomic disparities in PNC coverage demands collective responsibility and coordinated action from policymakers, healthcare providers, and communities globally.

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 Hamadan University of Medical Sciences approved this study with the code of IR.UMSHA.REC.1401.265. 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

AM: Conceptualization, Formal analysis, Writing – original draft, Writing – review & editing. FS: Conceptualization, Writing – original draft, Writing – review & editing. MA: Data curation, Methodology,

Writing – original draft, Writing – review & editing. RB: Investigation, 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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Barriers to care for musculoskeletal sarcoma patients: a public health perspective

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Introduction: This study seeks to investigate the barriers to care that exist for patients presenting with sarcomas of musculoskeletal origin. Understanding the roots of delays in care for patients with musculoskeletal sarcoma is particularly important given the necessity of prompt treatment for oncologic diagnoses. Investigators reviewed relevant studies of publications reporting barriers to care in patients undergoing diagnosis and treatment of musculoskeletal tumors.

Methods: A comprehensive literature search was conducted using Scopus, Embase, Web of Science, and PubMed-MEDLINE. Twenty publications were analyzed, including a total of 114,056 patients.

Results: Four barrier subtypes were identified: Socioeconomic Status, Geographic Location, Healthcare Quality, Sociocultural Factors. Socioeconomic status included access to health insurance and income level. Geographic location included distance traveled by patients, access to referral centers, type of hospital system and resource-challenged environments. Healthcare quality included substandard imaging, access to healthcare resources, and healthcare utilization prior to diagnosis. Sociocultural factors included psychological states, nutrition, education and social support.

Conclusion: After identifying the most significant barriers in this study, we can target specific public health issues within our community that may reduce delays in care. The assessment of barriers to care is an important first step for improving the delivery of oncologic patient care to this patient population.

KEYWORDS

access barriers in oncology, socioeconomic determinants of cancer care, public health, geographic barriers to care, musculoskeletal sarcoma, delays in care

1 Introduction

Sarcomas are malignancies of the body's connective tissue. They represent a subset of primary musculoskeletal cancers that account for less than 1% of all diagnosed cancers annually (1). These tumors are associated with significant morbidity and mortality and require prompt diagnosis and highly specialized treatment to achieve favorable outcomes (1). However, disparities in access to resources and inequitable distribution of care can create

significant obstacles, resulting in delays in the delivery of healthcare services (2–4).

Numerous factors exist that may play a role in delayed patient presentation and can be broadly categorized by the five social determinants of health: economic stability, health care and quality, social and community context, neighborhood and built environment, and education (5, 6). These overlapping categories provide a context for understanding the barriers to care that exist in individuals with bone sarcoma. The authors believe that an in-depth understanding of the barriers to care for patients with bone sarcoma is particularly important given that without timely interventions, the disease may quickly progress beyond the limits of treatment (3, 4).

To the author's knowledge, a systematic review of the barriers to care encountered by patients with musculoskeletal sarcoma has not been performed. Therefore, the aim of this study is to perform a high-quality systematic review of the literature to determine the most significant barriers to care that exist for patients undergoing diagnosis and treatment of musculoskeletal sarcoma.

2 Methods

2.1 Data source

Two experienced authors (HR, MB) conducted a comprehensive literature search using Scopus, Embase, Web of Science, and PubMed-MEDLINE databases from inception to June 6th, 2023. The search aimed to identify cohort studies, prospective and retrospective trials, randomized controlled trials, quasi-randomized control trials, and case series focusing on barriers to care for musculoskeletal sarcoma. The literature screening process was completed in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Supplementary Figure 1). Database search terms included specific search phrases. Use of Boolean operators were as follows: (sarcoma) AND (orthopedics OR orthopedics OR musculoskeletal) AND (barriers OR obstacles OR challenges OR difficulties). Minor adjustments were made to the search phrase to accommodate the different databases. Any discrepancies were settled following the presentation to a senior author (JA; Supplementary Figure 1).

2.2 Study selection

Inclusion criteria were applied as follows: (1) full-text accessible; (2) published in English; (3) inclusion of multiple patients; (4) analyzed primary data (5) level 4 evidence or above; (6) investigation of barriers to care for musculoskeletal tumors. Exclusion criteria encompassed: (1) systematic reviews and meta-analyses; (2) individual case studies; (3) management of solely non-orthopedic cancers.

Initially, abstracts and titles were screened to retain only studies of musculoskeletal sarcomas which identified a barrier to optimal care for patients in diagnosis and/or treatment. Our definition of musculoskeletal sarcoma includes primary sarcoma of the bone, soft tissue, or connective tissue origin within the musculoskeletal system. Subsequently, two reviewers (HR, MB) independently extracted relevant data from each included full-text study such as author and publication year, study type, musculoskeletal tumor diagnosis, type of barrier to care, treatment course, and clinical outcomes if available (Table 1).

2.3 Data extraction

No advanced statistical analyses could be performed due to the heterogeneity of study populations and differences in inquiry regarding specific barriers to care among the selected manuscripts.

3 Results

From a total of 1,504 publications initially screened, 20 articles met the eligibility criteria and were included in the final analysis. These articles comprised a collective sample of 114,056 patients. The included articles were further categorized into four distinct barrier subtypes as defined by the authors: socioeconomic status, geographic location, healthcare quality, and sociocultural factors. Five articles met the inclusion criteria for each barrier subtype.

3.1 Socioeconomic status

Five articles (25%) reported on barriers relating to socioeconomic barriers in musculoskeletal sarcoma care (Table 2).

Among the articles, four focused on the impact of insurance on musculoskeletal sarcoma care. Miller et al. (7) found that Medicaid insurance was associated with a hazard ratio (HR) of 1.18, indicating reduced survival rates in various types of sarcomas when compared to private insurance (7). Similarly, Smartt et al. reported HR values of 1.3 ($p = 0.003$) for osteosarcoma and 1.2 ($p = 0.019$) for soft tissue sarcoma for Medicaid insurance, further supporting the association with poorer outcomes (8). Consistent with these findings, Jang et al. revealed a HR of 1.28 ($p = 0.026$) for Medicaid insurance, indicating a higher mortality rate (3). Additionally, uninsured patients faced poorer outcomes when compared to patients with non-medicaid insurance, as observed in Smartt et al. study (HR 1.6, $p = 0.001$) (8).

Furthermore, Malik et al. explored the effect of insurance legislation on cancer staging and identified a positive impact following Medicaid expansion, with an increase in early-stage primary bone sarcomas ($p < 0.001$) and a decrease in late-stage cancers ($p < 0.001$) (9).

In terms of socioeconomic factors, Monsereenusorn et al. investigated treatment refusal and abandonment in osteosarcoma patients and discovered that lower income classifications in Southeast Asian countries were associated with higher rates of such occurrences ($p < 0.002$ for Philippines and $p < 0.025$ for Singapore) (3). Additionally, Miller et al. (7) also found that the lowest quartile Socioeconomic Status (SES) was a predictor of decreased 5-year survival (HR 1.23) (7).

3.2 Geographic location

Five articles (25%) examined barriers related to geography or locational factors in the context of musculoskeletal sarcoma care (Table 3). These studies explored the impact of access to regional referral centers, geographic regions, distance traveled, and

TABLE 1 Cited sarcoma studies: year, article type, journal, country, sample size, and cancer diagnosis.

Authors (Year)	Title	Article type	Journal	Country of origin	Sample size	Cancer diagnosis
Miller et al. (2017)	Socioeconomic measures influence survival in osteosarcoma: an analysis of the National Cancer Data Base	Retrospective Review	The International Journal of Cancer Epidemiology, Detection, and Prevention	USA	$N = 3{,}505$ $n_{Medicaid} = 869$ $n_{lowest\ quartile\ SES} = 683$	High-grade conventional osteosarcoma
Smartt et al. (2020)	Is There an Association Between Insurance Status and Survival and Treatment of Primary Bone and Extremity Soft-tissue Sarcomas? A SEER Database Study	Retrospective Review	Clinical Orthopedics and Related Research	USA	$N_{osteosarcoma} = 4{,}144$ $n_{non\text{-}Medicaid} = 3{,}098$ $n_{Medicaid} = 884$ $n_{uninsured} = 162$ $N_{soft\ tissue\ sarcoma} = 7{,}508$ $n_{non\text{-}Medicaid} = 6{,}292$ $n_{Medicaid} = 904$ $n_{uninsured} = 312$	Bone and soft tissue sarcomas
Jang et al. (2023)	Effect of Insurance Status on Mortality in Adults With Sarcoma of the Extremities and Pelvis: A SEER-Medicare Study	Retrospective Review	Journal of the American Academy of Orthopedic Surgeons	USA	$N = 7{,}056$ $n_{medicaid} = 182$ $n_{uninsured} = 34$ $n_{private\ ins} = 1{,}533$ $n_{medicare} = 4{,}785$	Bone and soft tissue sarcomas
Malik et al. (2021)	Has the Affordable Care Act Been Associated with Increased Insurance Coverage and Early-stage Diagnoses of Bone and Soft-tissue Sarcomas in Adults?	Retrospective Review	Clinical Orthopedics and Related Research	USA	$N = 15{,}287$ $n_{pre\text{-}ACA} = 6{,}537$ $n_{pre\text{-}Medicaid\ expansion} = 5{,}076$ $n_{post\text{-}Medicaid\ expansion} = 3{,}674$	Primary malignant bone tumors
Monsereenusorn et al. (2022)	Impact of treatment refusal and abandonment on survival outcomes in pediatric osteosarcoma in Southeast Asia: A multicenter study	Retrospective Review	Pediatric Blood & Cancer	Thailand	$N = 208$ $n_{TxRA} = 59$ $n_{non\text{-}TxRA} = 149$	Osteosarcoma
Wendt et al. (2019)	Rural patients are at risk for increased stage at presentation and diminished overall survival in osteosarcoma	Retrospective Review	Cancer Epidemiology	USA	$N = 476$ $n_{>2\ h\ drive} = 128$	High Grade osteosarcoma
Fayet et al. (2022)	No Geographical Inequalities in Survival for Sarcoma Patients in France: A Reference Networks' Outcome?	Retrospective Review	Cancers	France	$N = 2{,}281$ $n_{wealthy\ metropolitan\ area} = 468$ $n_{precarious\ district} = 1{,}188$	All Sarcomas: 6.8% bone, 69% soft-tissue, and 14.2% visceral sarcomas
Fayet et al. (2021)	Determinants of the access to remote specialized services provided by national sarcoma reference centers	Retrospective Review	BMC Cancer	France	$N = 20{,}589$	All Sarcomas: orthopedic and non-orthopedic sarcomas
Fujiwara et al. (2021)	Greater travel distance to specialized facilities is associated with higher survival for patients with soft-tissue sarcoma: US nationwide patterns	Retrospective Review	PLOS One	USA	$N = 34{,}528$ $n_{>101\ miles} = 2{,}143$ $n_{51\text{-}100\ miles} = 2{,}761$ $n_{11\text{-}50\ miles} = 12{,}729$ $n_{<10\ miles} = 16{,}895$	Soft tissue sarcoma
Sasi et al. (2023)	Determinants and impact of diagnostic interval in bone sarcomas: A retrospective cohort study	Retrospective Review	Pediatric Blood & Cancer	India	$N = 1{,}227$ $n_{osteosarcoma} = 470$ $n_{ewing\ sarcoma} = 757$	High grade Osteosarcoma, Ewing Sarcoma

(Continued)

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TABLE 1 (Continued)

Authors (Year)	Title	Article type	Journal	Country of origin	Sample size	Cancer diagnosis
Rædkjær et al. (2019)	Use of Healthcare Services Two Years before Diagnosis in Danish Sarcoma Patients, 2000–2013	Retrospective cohort study	Sarcoma	Denmark	$N = 2{,}167$ $n_{female} = 972$ $n_{male} = 1{,}195$ $N_{Reference\ Matched\ Cohort} = 21{,}670$	Soft tissue sarcoma and osteosarcoma
Schiavi et al. (2015)	Using a family history questionnaire to identify adult patients with increased genetic risk for sarcoma	Self Administered Sarcoma Clinic Genetic Screening (SCGS)	Current Oncology	Canada	$N = 164$ $n_{female} = 102$ $n_{female} = 62$	All Sarcomas
Weaver et al. (2020)	The complexity of diagnosing sarcoma in a timely manner: perspectives of health professionals, patients, and carers in Australia	Exploratory Qualitative Research Design	BMC Health Service Research	Australia	$N = 60$ $n_{health\ prof\ working\ w/\ sarcoma\ pts} = 21$ $n_{pts\ diagnosed\ w/\ sarcoma} = 22$ $n_{caregivers\ for\ ppl\ diagnosed\ w/\ sarcoma} = 17$	Sarcoma
Dahan et al. (2017)	Proximal femoral osteosarcoma: Diagnostic challenges translate into delayed and inappropriate management	Retrospective Review	Orthopedics & Traumatology: Surgery & Research	France	$N = 12$ $n_{incorrect\ imaging} = 2$	Proximal femur osteosarcoma
Poudel et al. (2017)	Factors associated with local recurrence in operated osteosarcomas: A retrospective evaluation of 95 cases from a tertiary care center in a resource challenged environment	Retrospective Review	Journal of Surgical Oncology	India	$N = 95$ $n_{local\ recurrence} = 15$ $n_{no\ local\ recurrence} = 80$	Osteosarcoma
Hewitt et al. (2019)	Patient Perceptions of the Impact of Treatment (Surgery and Radiotherapy) for Soft Tissue Sarcoma	Single, Semi-structured Interviews	Sarcoma	United Kingdom	$N = 19$	Soft tissue sarcoma
Sasaki et al. (2018)	Validation of Different Nutritional Assessment Tools in Predicting Prognosis of Patients with Soft Tissue Spindle-Cell Sarcomas	Retrospective Review	Nutrients	Japan	$N = 103$ $n_{death\ within\ 1\ yr} = 15$ $n_{1\ yr\ survival} = 88$	Soft tissue spindle-cell sarcomas
Alamanda et al. (2014)	Effect of marital status on treatment and survival of extremity soft tissue sarcoma	Retrospective Review	Annals of Oncology	USA	$N = 7{,}384$ $n_{single} = 2{,}977$	Soft tissue sarcoma
Alamanda et al. (2015)	Racial Disparities in Extremity Soft-Tissue Sarcoma Outcomes A Nationwide Analysis	Retrospective Review	American Journal of Clinical Oncology	USA	$N = 7{,}225$ $n_{African\ American} = 825$	Soft tissue sarcoma
Siddiqui et al. (2015)	Neglected orthopedic oncology--Causes, epidemiology and challenges for management in developing countries	Retrospective Review	Indian Journal of Cancer	India	$N = 18$ $n_{low\ SES} = 15$ $n_{uneducated} = 17$	Bone and soft tissue sarcomas

resource-challenged environments on diagnosis, outcomes, and patient experiences.

Wendt et al. revealed that patients with over a 2-h drive to comprehensive cancer centers had increased incidence of metastases and higher mortality rates ($p = 0.021$) (10). Rural status was also associated with increased mortality, independent of tumor size (HR 1.58, $p = 0.037$). Additionally, Sasi et al. explored the impact of resource-challenged environments on diagnostic intervals and

TABLE 2 Insights into socioeconomic barriers from five key studies.

Study	Identified barrier	Sample size	Reported outcomes
Miller et al., 2017 (Cancer Epidemiol)	Insurance status & SES	$N = 3{,}505$ $n_{Medicaid} = 869$ $n_{lowest\ quartile\ SES} = 683$	Lowest quartile SES (HR 1.23) & Medicaid insurance (HR 1.18) predictors of decreased survival at 5 years
Smartt et al., 2020 (Clin Orthop Relat Res)	Insurance status	$N_{osteosarcoma} = 4{,}144$ $n_{non\text{-}Medicaid} = 3{,}098$ $n_{Medicaid} = 884$ $n_{uninsured} = 162$ $N_{soft\ tissue\ sarcoma} = 7{,}508$ $n_{non\text{-}Medicaid} = 6{,}292$ $n_{Medicaid} = 904$ $n_{uninsured} = 312$	Medicaid insurance had reduced survival than did patients with non-Medicaid insurance in both osteosarcoma and soft tissue sarcoma (HR 1.3, $p = 0.003$ & HR 1.2, $p = 0.019$) Uninsured patients had reduced survival with extremity soft-tissue sarcomas (HR 1.6, $p = 0.001$)
Jang et al., 2023 (J Am Acad Orthop Surg)	Insurance status	$N = 7{,}056$ $n_{medicaid} = 182$ $n_{uninsured} = 34$ $n_{private\ ins} = 1{,}533$ $n_{medicare} = 4{,}785$	Medicaid insurance as the primary insurer had a 28% higher mortality compared to private insurance (HR 1.28, $p = 0.026$) Medicare as the primary insurer showed no significant difference in mortality compared to private insurance (HR, 1.06, $p = 0.243$)
Malik et al., 2021 (Clin Orthop Relat Res)	Insurance legislation	$N = 15{,}287$ $n_{pre\text{-}ACA} = 6{,}537$ $n_{pre\text{-}Medicaid\ expansion} = 5{,}076$ $n_{post\text{-}Medicaid\ expansion} = 3{,}674$	Post Medicaid expansion, the proportion of early stage primary bone sarcomas increased ($p < 0.001$), and the proportions of late-stage cancers decreased ($p < 0.001$)
Monsereenusorn et al., 2022 (Pediatr Blood Cancer)	Predictive factors (SES) for treatment refusal and abandonment (TxRA)	$N = 208$ $n_{TxRA} = 59$ $n_{non\text{-}TxRA} = 149$	Income classification of countries in Southeast Asia (Philippines: $p < 0.002$, Singapore: $p < 0.025$) was associated with greater TxRA

TABLE 3 Exploring geographic barriers in musculoskeletal sarcoma care: findings from five studies.

Study	Identified barrier	Sample size	Reported outcomes
Wendt et al., (Cancer Epidemiol)	Access to regional referral centers	$N = 476$ $n_{>2\ h\ drive} = 128$ did not describe how many counted as rural	Patients with >2-h drive to comprehensive cancer center showed increased incidence of metastases ($p = 0.021$) Rural status associated with increased mortality when controlling for size of the tumor (HR 1.58, $p = 0.037$)
Fayet et al., 2022 (Cancers)	Geographic region	$N = 2{,}281$ $n_{wealthy\ metropolitan\ area} = 468$ $n_{precarious\ district} = 1{,}188$	Precarious population districts associated with lower survival (HR 1.23) however no significant association in survival after adjustment for the clinical variables (HR 1.03)
Fayet et al., 2021 (BMC Cancer)	Rural region & social deprivation	$N = 20{,}589$	Patients who are the farthest from reference centers have a reduced likelihood of early access to specialized diagnosis [OR 1.18, 95% CI 1.06 to 1.31] and MTB discussion [OR 1.24, 95% CI 1.10 to 1.40]. However, the impact of distance is relatively small compared to clinical factors and previous research on accessing cancer-specialized facilities.
Fujiwara et al., 2021 (PLoS One)	Distance traveled to academic/research center	$N = 34{,}528$ $n_{>101\ miles} = 2{,}143$ $n_{51\text{-}100\ miles} = 2{,}761$ $n_{11\text{-}50\ miles} = 12{,}729$ $n_{<10\ miles} = 16{,}895$	Distance >100 miles traveled associated with greater overall survival compared to distance <10 miles traveled (HR 0.877, $p < 0.001$) Diagnosis at an academic/research institution associated with greater overall survival (HR 0.857)
Sasi et al., 2023 (Pediatr Blood Cancer)	Resource-challenged environment	$N = 1{,}227$ $n_{osteosarcoma} = 470$ $n_{ewing\ sarcoma} = 757$	Distance greater than 100 km was a predictor of a longer diagnostic interval (>4 months; $p < 0.04$) Place of residence did not impact the proportion of patients with good necrosis post neoadjuvant chemotherapy ($p = 0.30$)

treatment outcomes in high-grade osteosarcoma and Ewing sarcoma patients. They discovered that a distance greater than 100 km was a predictor of a longer diagnostic interval ($p < 0.04$), while place of residence did not significantly impact the proportion of patients with good necrosis post neoadjuvant chemotherapy ($p = 0.30$) (11).

Contrasting results were found when Fujiwara et al. investigated the effect of distance traveled to academic/research centers on overall

survival in soft tissue sarcoma patients (12). Their findings revealed that traveling more than 100 miles was associated with greater overall survival (HR 0.877, $p < 0.001$), and diagnosis at an academic/research institution was also linked to improved outcomes (HR 0.857).

Fayet et al. investigated the influence of geographic regions on sarcoma survival. They found that living in precarious population districts was initially associated with lower survival rates (HR 1.23), but this association became non-significant after adjusting for clinical variables (HR 1.03). Another study by Fayet et al. explored the impact of rural regions and social deprivation on access to specialized diagnosis [Odds Ratio (OR) 1.18, 95% Confidence Interval (CI) 1.06 to 1.31] and multidisciplinary tumor board (MTB) discussions [OR 1.24, 95% CI 1.10 to 1.40] (13). Patients residing farther from reference centers had reduced likelihoods of early access to specialized diagnosis and MTB discussions. However, the influence of distance was relatively small compared to clinical factors and previous research on accessing cancer-specialized facilities.

3.3 Healthcare quality

Five articles (25%) examined barriers relevant to the diagnostic period of musculoskeletal sarcomas (Table 4). These studies focused on various factors impacting timely diagnosis, including healthcare utilization prior to diagnosis, access to genetic questionnaires, delays in diagnosis, substandard imaging, and resource-challenged environments.

Rædkjær et al. and Weaver et al. both identified delays in diagnosis as a significant barrier. Rædkjær et al. emphasized the increased utilization of healthcare services by sarcoma patients leading up to their diagnoses. They found that sarcoma patients had significantly higher incidence rate ratios of healthcare service use compared to a matched cohort during the year leading up to their diagnoses. The highest incidence rate ratio was observed in the last month prior to diagnosis with an IRR of 13.89 (CI 12.41–15.54) (14). Weaver et al. conducted exploratory qualitative research on factors contributing to delays in sarcoma diagnosis (4). Limited availability of health services, lack of prompt referrals to sarcoma specialists, and diagnostic challenges were identified as barriers associated with delays in diagnosis.

Schiavi et al. and Dahan et al. focused on specific aspects of the diagnostic process. Schiavi et al. identified that a family history of cancer (up to 3rd-degree relatives) was reported in 69% patients diagnosed with a sarcoma, and highlighted the importance of access to genetic questionnaires in identifying patients who may benefit from genetic assessment (15). On the other hand, Dahan et al. discussed the consequences of substandard imaging practices and how 1 of the 2 patients who underwent inappropriate imaging experienced a local recurrence and metastases after 6 years and died 1 year later (16).

Poudel et al. observed that patients who had undergone a previous biopsy procedure outside of the home institution had a higher

TABLE 4 Insights into diagnostic barriers in musculoskeletal sarcoma: key findings from five studies.

Study	Identified barrier	Sample size	Reported outcomes
Rædkjær et al., 2019 (Sarcoma)	Utilization of healthcare prior to diagnosis	$N = 2{,}167$ $n_{female} = 972$ $n_{male} = 1{,}195$ $N_{Reference\ Matched\ Cohort} = 21{,}670$	Sarcoma patients had significantly increased incidence rate ratios (IRR) in use of healthcare services compared to the matched cohort a year before their diagnoses. The IRRs were statistically significant during the 12 months leading up to the diagnosis, reaching its highest point in the last month with an IRR of 13.89 (CI 12.41–15.54). No significant differences in length of increased consultation rates between sarcoma type, stage, and grade
Schiavi et al., 2015 (Curr Oncol)	Access to genetic questionnaires	$N = 164$ $n_{female} = 102$ $n_{female} = 62$	A family history of cancer (up to 3rd-degree relatives) was reported in 69% patients diagnosed with a sarcoma. The SCGS questionnaire was valuable in identifying sarcoma patients who may benefit from a genetic assessment. By using this tool, one can identify families who meet the criteria for LFL gene evaluation.
Weaver et al., 2020 (BMC Health Serv Res)	Delay of Diagnosis	$N = 60$ $n_{health\ prof\ working\ w/\ sarcoma\ pts} = 21$ $n_{pts\ diagnosed\ w/\ sarcoma} = 22$ $n_{caregivers\ for\ ppl\ diagnosed\ w/\ sarcoma} = 17$	Delays in diagnosis were associated with the limited availability of health services, lack of prompt referrals to a sarcoma specialist, and diagnostic challenges
Dahan et al., 2017 (Orthop Traumatol Surg Res)	Substandard imaging	$N = 12$ $n_{incorrect\ imaging} = 2$	Management was inappropriate in 2 (17%) patients. Patients did not undergo all the recommended imaging studies prior to surgery. Subsequently 1 of the 2 patients experienced a local recurrence and metastases after 6 years and died 1 year later.
Poudel et al., 2017 (J Surg Oncol)	Resource challenged environments of the developing world	$N = 95$ $n_{local\ recurrence} = 15$ $n_{no\ local\ recurrence} = 80$	More patients who had undergone a previous biopsy procedure outside of the home institution were found to have local recurrence (LR) compare to no local recurrence (NLR; $p = 0.05$) The mean delay in biopsy of the NLR group was 4.16 ± 4.81 weeks compared to 9.46 ± 6.5 weeks in the LR group ($p = 0.0002$)

incidence of local recurrence ($p = 0.05$) (17). Additionally, the mean delay in biopsy was significantly longer in the group with local recurrence compared to the group with no local recurrence ($p = 0.0002$).

3.4 Sociocultural factors

Five articles (25%) examined sociocultural barriers to care in patients with musculoskeletal sarcoma (Table 5). These studies shed light on various aspects of sociocultural factors that influence the patient experience and outcomes. Among these articles, one study specifically focused on how psychological states and mental health could manifest as barriers to care.

Hewitt et al. conducted single, semi-structured interviews with patients diagnosed with soft tissue sarcoma (18). The interviews revealed that concerns throughout treatment included a lack of understanding of soft tissue sarcomas and apprehension about treatment plans. The study suggested that implementing social support networks could make treatment more bearable for patients.

Sasaki et al. conducted a retrospective review of patients with soft tissue spindle-cell sarcomas (19). Their findings highlighted the impact of access to nutrition on patient outcomes. Patients with a higher Glasgow Prognostic Score, ($p < 0.001$), Geriatric Nutritional Risk Index ($p < 0.001$), and controlling nutritional (CONUT) score ($p < 0.001$) had a significantly higher risk of death within 1 year of diagnosis. The study emphasized the importance of adequate nutrition in improving prognosis.

Alamanda et al. and Alamanda et al. both focused on sociocultural factors such as marital status and race in relation to the diagnosis and treatment of soft tissue sarcomas (20, 21). Alamanda et al. found that being single was associated with higher grade tumors ($p = 0.013$), less radiotherapy ($p < 0.001$), and fewer surgeries ($p < 0.001$). Single status was also identified as an independent predictor of sarcoma-specific death ($p < 0.0001$). In Alamanda et al.'s study, African American race was associated with larger tumor size ($p < 0.001$), less radiotherapy ($p = 0.024$), fewer surgeries ($p = 0.002$), and greater number of deaths ($p < 0.001$).

Siddiqui et al. explored the influence of education status and SES on the delay in seeking medical care among patients with bone and soft tissue sarcomas (22). Patients with low education levels and low SES experienced delays in seeking medical care due to financial constraints, cultural and religious beliefs, and lack of access to healthcare facilities.

4 Discussion

The present study documents the types of barriers to care encountered by musculoskeletal sarcoma patients. To the authors' knowledge, this is the first study to systematically assess barriers to care by socioeconomic status, geographic location, healthcare quality, and sociocultural factors.

The major theme of these results emphasizes how low socioeconomic status (SES) constitutes the underlying common denominator and the most important barrier to care for this patient population. A multivariate analysis of the 2022 National Cancer Database demonstrated greater mortality of uninsured sarcoma patients in the first 2 years when compared to their insured counterparts (23). The association between insurance status and increased mortality was unchanged after adjusting for potential confounders, including disease stage at presentation (3, 24). Prior to the expansion of Medicaid in 2014 the mortality of uninsured sarcoma patients was 28% greater than their insured counterparts. Since the Medicaid expansion, we have seen a decrease in sarcoma mortality demonstrating the direct link between increased access to care and survivorship (3). This relationship may be partially explained by the fact that insurance status has been demonstrated to be a positive predictor of clinic

TABLE 5 Sociocultural barriers in musculoskeletal sarcoma care: insights from five key studies.

Study	Identified barrier	Sample size	Reported outcomes
Hewitt et al., 2019 (Sarcoma)	Psychological	$N = 19$	Interviews showed that concerns throughout treatment included lack of understanding of soft tissue sarcomas and apprehension about treatment plans. Further interviewers concluded that treatment could be perceived as being more bearable if social support networks are implemented
Sasaki et al., 2018 (Nutrients)	Access to nutrition	$N = 103$ $n_{\text{death within 1 yr}} = 15$ $n_{\text{1 yr survival}} = 88$	Higher Glasgow Prognostic Score, ($p < 0.001$), Geriatric Nutritional Risk Index ($p < 0.001$), and controlling nutritional (CONUT) score ($p < 0.001$) significantly differed between patients who died within 1 year and patients who lived longer. Higher Glasgow Prognostic Score ($p < 0.004$) is a risk factor for death within a year of diagnosis.
Alamanda et al., 2014 (Ann Oncol)	Marital status	$N = 7{,}384$ $n_{\text{single}} = 2{,}977$	Single status was associated with higher grade tumors ($p = 0.013$), less radiotherapy ($p < 0.001$), and fewer surgeries ($p < 0.001$). Single status found to be an independent predictor of sarcoma specific death ($p < 0.0001$)
Alamanda et al., 2015 (Am J Clin Oncol)	Race	$N = 7{,}225$ $n_{\text{African American}} = 825$	African American race was associated with larger tumor size ($p < 0.001$), less radiotherapy ($p = 0.024$), fewer surgeries ($p = 0.002$), and greater number of deaths ($p < 0.001$)
Siddiqui et al., 2015 (Indian J Cancer)	Education Status & SES	$N = 18$ $n_{\text{low SES}} = 15$ $n_{\text{uneducated}} = 17$	Causes of delay in seeking medical by patients of low education level and low SES was attributed to financial constraints, cultural and religious believes, and lack of access to health care facilities

attendance which can prevent delays in care and advanced-stage presentation of sarcoma (25, 26).

An additional barrier to care resulting in delays in diagnosis is the concept of distance decay in which the outcomes of oncology patients decrease the farther away they live from a referral center, as long-distance was associated with increased wait time for diagnostic investigations (25). Our study demonstrated that prolonged distance and rural status were associated with increased morbidity and mortality in some cases, but not all (27). The reasons for these contradictory findings are multifaceted and may include other confounders that lead to delays in diagnosis.

Delays in diagnosis are identified as a significant barrier, with factors such as limited availability of health services, lack of prompt referrals to specialists, and diagnostic challenges contributing to these delays. Substandard imaging practices were also highlighted as a concern, with adverse outcomes observed in patients who did not undergo recommended imaging studies before surgery. Access to genetic questionnaires was identified as crucial for identifying patients who may benefit from a genetic assessment. The studies conducted in resource-challenged environments further emphasized the impact of such settings on both the quality of care and outcomes for sarcoma patients.

Previous literature provides insights into the current management of bone sarcomas. Gutowski et al. emphasize the role of chemotherapy advancements in improving survival for bone sarcoma patients (28). Similarly, Böhm et al. also discuss differentiated treatment approaches for malignant primary bone tumors, such as osteosarcoma, chondrosarcoma, and Ewing's sarcoma, which have shown notable improvements with adjuvant chemotherapy (29). If patients encounter barriers such as diagnostic delays, substandard imaging, limited access to genetic questionnaires, or resource-challenged environments, they may face difficulties in receiving these standardized treatments, resulting in poorer outcomes.

The results of our review corroborate previous literature which focused on the effect of healthcare quality on cancer outcomes. Moor et al. emphasized the significance of access to both cancer and general medical care for cancer survivors, by highlighting that survivors who did not receive necessary cancer care had lower education levels and higher rates of public or no insurance compared to those who received all required care (30). Arhi et al. demonstrated that delays in referral from primary care resulted in later-stage colorectal cancer diagnosis and worse prognosis (31). Furthermore, Aparicio et al. reveal that a substantial proportion (52%) of older adult patients with colorectal cancer receive sub-standard treatment (32). These results are similar to results we found in sarcoma with difficulty in diagnosis and sub-optimal imaging.

The barriers to care identified during the diagnostic period of musculoskeletal sarcoma include delays in diagnosis, substandard imaging practices, limited access to genetic questionnaires, and the impact of resource-challenged environments. Addressing these barriers is crucial to improving healthcare quality for sarcoma patients. Further research and improvements in healthcare systems are warranted to ensure timely diagnosis, appropriate imaging practices, and access to genetic assessment, particularly in resource-challenged environments.

The findings on sociocultural factors in musculoskeletal sarcoma care reveal the significance of addressing patient concerns, implementing social support networks, and ensuring access to adequate nutrition. These factors contribute to improved treatment experiences and patient outcomes. The influence of sociocultural factors such as marital status and race is evident, with single status associated with higher-grade tumors and poorer treatment outcomes, while African American race is linked to larger tumor size and increased mortality. Additionally, education status and socioeconomic status impact delays in seeking medical care due to various barriers. These findings highlight the importance of addressing sociocultural barriers to enhance sarcoma care and optimize patient outcomes.

The results from our review corroborate the existing literature underscoring the significance of sociocultural factors in cancer care. Haier et al. emphasize the implementation and modification of cancer care systems, particularly in low-and middle-income countries (LMICs) (33). Understanding and utilizing sociocultural incentives, such as free housing and access to education, are crucial in addressing resource challenges and improving care, especially for vulnerable populations like metastatic cancer patients in LMICs. Ward et al. highlight disparities in cancer outcomes related to race/ethnicity and SES, with residents of poorer counties having higher death rates from cancer (34). Additionally, even when accounting for poverty rates, African American, American Indian/Alaskan Native, Asian/Pacific Islander men, and African American and American Indian/Alaskan Native women, had lower five-year survival rates compared to non-Hispanic Whites. Alcindor et al. emphasize the importance of multidisciplinary team care at expert centers for sarcoma treatment (25). They report significantly improved oncologic outcomes for patients treated at high-volume centers. This finding suggests that access to specialized care and expertise plays a critical role in improving sarcoma treatment outcomes. Valencia et al. observe excessive mortality risk among BIPOC (Black, Indigenous, and People of Color) individuals compared to non-Hispanic White counterparts and note disparities in engagement in routine cancer screenings, treatment initiation, surgical interventions, and higher mortality rates within 5 years of diagnosis (35).

Although this paper provides an enhanced understanding to barriers to care in patients diagnosed with musculoskeletal sarcoma, the present study is not without limitations. As with many systematic reviews, this study is susceptible to biases including publication and selection biases. Further, there was a lack of homogeneity in the reporting of variables including healthcare quality measures, healthcare costs, sociocultural demographics, and geographic measures. This is likely secondary to the lack of prospective studies on the topic. For this reason, we were unable to perform high power statistical analyses, limiting the ability to critically appraise and draw comparisons from the published studies to date. In spite of the aforementioned shortcomings, the present study is the first to provide a comprehensive summary and evaluation of the most pressing barriers to diagnose and treatment of sarcomas of musculoskeletal origin. Future researchers should be focused on developing measurement tools and questionnaires that are widely accessible to more efficaciously capture factors that influence the care of patients with sarcomas of musculoskeletal origin.

The findings on sociocultural factors in musculoskeletal sarcoma care provide insights into the patient experience and outcomes. The identified barriers to care call attention to the need for social support networks, secure access to adequate nutrition, address disparities based on marital status and race, and improve healthcare access for individuals with low education levels and low SES. By addressing these

barriers, healthcare systems can strive for more equitable care and enhance treatment outcomes for patients with sarcoma.

5 Conclusion

Given the variety of barriers that exist for patients with musculoskeletal sarcoma, key initiatives related to increasing accessibility to care within specific patient communities may reduce delays to care for oncologic patients. These barriers to care highlight the importance of public health initiatives focused on improving patient access based on both internal and external patient factors. Further studies are warranted to explore specific interventions that improve patient access and prevent sarcoma progression to an untreatable or complex surgical stage.

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

AS: Conceptualization, Data curation, Formal analysis, Methodology, Software, Writing – original draft, Writing – review & editing. MB: Data curation, Investigation, Writing – original draft, Writing – review & editing. JA: Conceptualization, Supervision, Writing – original draft, Writing – review & editing. HR: Data curation, Writing – original draft, Writing – review & editing, Investigation. MH: Supervision, Writing – review & editing. BC: 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

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1399471/full#supplementary-material

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Developing methods to identify resilience and improve communication about diagnosis in pediatric primary care

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Communication underlies every stage of the diagnostic process. The Dialog Study aims to characterize the pediatric diagnostic journey, focusing on communication as a source of resilience, in order to ultimately develop and test the efficacy of a structured patient-centered communication intervention in improving outpatient diagnostic safety. In this manuscript, we will describe protocols, data collection instruments, methods, analytic approaches, and theoretical frameworks to be used in to characterize the patient journey in the Dialog Study. Our approach to characterization of the patient journey will attend to patient and structural factors, like race and racism, and language and language access, before developing interventions. Our mixed-methods approach is informed by the Systems Engineering Initiative for Patient Safety (SEIPS) 3.0 framework (which describes the sociotechnical system underpinning diagnoses within the broader context of multiple interactions with different care settings over time) and the Safety II framework (which seeks to understand successful and unsuccessful adaptations to ongoing changes in demand and capacity within the healthcare system). We will assess the validity of different methods to detect diagnostic errors along the diagnostic journey. In doing so, we will emphasize the importance of viewing the diagnostic process as the product of communications situated in systems-of-work that are constantly adapting to everyday challenges.

KEYWORDS

diagnosis, diagnostic process, patient safety, safety II, pediatrics, communication, primary care, ambulatory care

1 Introduction

Communication underlies every stage of the diagnostic process. As patients and families are present throughout the diagnostic process, effective communication with patients and families is both a definitional aspect of diagnostic excellence (1) and a pragmatic strategy for achieving it. Due to limitations of the current healthcare system, including fragmented outpatient care and limitations to access for patients who speak languages other than English, patients and families are often the ones communicating clinical information from one diagnostic encounter to another (2, 3). Research engaging families in safety reporting identified that families uncovered multiple diagnosis-related errors and adverse events, including delayed diagnoses of intussusception, aspiration pneumonia, and urosepsis (4, 5). The quality of bidirectional communication between patients, families/caregivers, and clinicians determines how diagnostic information is gathered, integrated into differential diagnoses, and communicated back to patients/caregivers, ultimately contributing to diagnostic outcomes. Yet, in spite of their important role in diagnostic communication, studies characterizing opportunities to improve diagnosis often lack the patient and family view (4, 6, 7).

In recognition of the critical shared role of patients, their families, and clinicians in diagnosis, the National Academy of Science, the National Quality Forum, the Institute for Healthcare Improvement, the US Center for Medicare & Medicaid Services, and the Children's Hospital Association have all called for improved patient engagement in the diagnostic process (1, 6, 8, 9). Engaging patients more closely in diagnostic and safety research is respectful, ethical, and ensures safer care (10–12). Robust communication between patients, families/caregivers, and clinicians is more likely to generate resilience and safeguard against diagnostic failures and harm. Conversely, communication failures contribute to nearly half of malpractice claims, with more than half being provider-patient communication failures (13). In particular, there is ample opportunity to improve effective communication about diagnostic uncertainty (14–17).

Vulnerable populations, including families from racial/ethnic minority groups, those with lower incomes, and those who speak languages other than English disproportionately experience communication failures in health care (7, 18). Disparities have been characterized in the diagnosis of varying conditions including depression, appendicitis, acute myocardial infarction, and breast cancer (19–23). It is particularly important to understand how pediatric diagnostic processes differ for families who speak languages other than English. For example, patients with limited English proficiency are four times as likely to report having no qualified provider or interpreter who spoke their language, three times as likely to report incorrect or out of date medical records, and twice as likely to report not understanding the follow-up plan (24).

Effective communication is imperative and challenging in pediatrics, where different parents and/or other caregivers often hand off responsibility for clinic appointments or childcare. Often, children have a limited ability to convey symptoms, relying on multiple family caregivers. This is particularly true among children with complex or disabling conditions who average 6.5 (SD 6.5) outpatient visits annually, most to primary care (25). Most of these children (60%) will see a specialist at least once a year, 20% will visit the emergency room, and 10% will be admitted to the hospital (25, 26).

Diagnostic errors have been defined by the National Academy of Medicine as, "the failure to (a) establish an accurate and timely explanation of the patient's health problem(s) or (b) communicate that explanation to the patients. This definition underscores this critical component that communication plays in avoiding delayed, missed, or wrong diagnoses. Opportunities to improve pediatric diagnosis, in terms of accuracy or communication, have not been well characterized (8). While an estimated 5% of adult primary care visits involve a diagnostic error (27), rates and characteristics of diagnostic errors are largely unquantified and undescribed in pediatric primary care. Research in specific disease conditions suggest that opportunities to improve pediatric diagnosis are common. Studies of hypertension or adolescent depression found that these diagnoses are missed 50% of the time in pediatric primary care (28). Up to 20% of children also have a delayed diagnosis of physical abuse (29–33). Diagnostic errors also occur with other common pediatric conditions, with misdiagnosis reported in 14% of asthma and 8% of appendicitis diagnoses (8). In a national study of pediatric health system leaders and parents, diagnostic safety was identified as a high priority topic for research (34). There is an urgent need to evaluate and develop interventions to improve diagnostic communication to prevent ongoing injury and death from diagnostic errors. This need is particularly urgent in primary care, where most of pediatric healthcare is delivered.

In this manuscript, we will describe the theoretical frameworks, protocols, data collection instruments, methods, and analytic approaches to be used in the Dialog Study. This study aims to characterize the pediatric diagnostic journey, focusing on communication as a source of resilience, in order to develop and adapt structured, patient-centered communication interventions for outpatient use, and to test the efficacy of such interventions in improving diagnostic safety. We will attend to patient and structural factors, like race and racism, and language and language access, before developing interventions.

2 Theoretical frameworks

Our mixed methods approach is grounded in the Systems Engineering Initiative for Patient Safety (SEIPS) 3.0 framework (35) which describes the sociotechnical system within the broader context of multiple clinical interactions that patients have in different care settings over time. We aimed to understand both the successful and unsuccessful adaptations to ongoing changes in demand and capacity within the healthcare system (36).

Recognizing that healthcare is "increasingly distributed over space and time," Carayon et al. recently updated her widely used Systems Engineering Initiative for Patient Safety (SEIPS) framework, to characterize the patient journey in multiple care settings over time (35). Generally, the SEIPS framework allows for analysis of patient outcomes as a product of the interaction between structures (people, environment, tasks, tools) and processes (37, 38). Grounding the understanding of diagnostic safety as a longitudinal journey constructed by the dynamic interplay of sociotechnical elements will guide our observations, simulations, and analysis.

Our study investigates the diagnostic journey through both a traditional "error" perspective (Safety I) and approaches to avert errors and achieve diagnostic excellence (Safety II). Identifying both opportunities to improve diagnosis and drivers of diagnostic

excellence is aligned with principles of "resilience engineering," which posit that safety is a consequence of adapting to the changing conditions of systems function (39). In other words, harm occurs not because an otherwise stable system malfunctioned but rather because inappropriate adaptive actions were taken within an ever-changing, inherently error-prone environment. Thus, resilient systems can (1) respond (*know what to do*), (2) monitor (*know what to look for*), (3) learn from experience, and (4) anticipate (*know what to expect*) (40). We will assess the presence and interplay of these factors and the role of robust communication in their presence along the diagnostic journey.

SEIPS 3.0 and Safety II align to overcome limitations of approaches that seek improvement by rectifying piecemeal individual errors to instead explore how resilient people and systems deliver appropriate diagnoses and what systems barriers and facilitators shape these processes. It allows for the prospective identification of the everyday practices that contribute to effective diagnosis, rather than relying on retrospective assessment of failures. Informed by these frameworks, we will assess the validity of different methods to detect diagnostic errors along the diagnostic journey. In doing so, we will extend the idea that the diagnostic process is the product of communications situated in systems-of-work that are constantly adapting to everyday challenges.

3 Methods and analysis

3.1 Study design

This is a prospective, mixed-methods, observational study aimed at characterizing opportunities to improve diagnosis and sources of systems resilience that drive diagnostic excellence for children with medical complexity presenting to primary care with acute concerns. We will adapt and test well-established patient safety research methods (including chart review, hospital incident reporting, family safety reporting, surveys, observations, and interviews) to characterize the diagnostic journey and associated successes and harms experienced by acutely ill children with multiple comorbid conditions and their families (Table 1). We will also apply reliable, valid, and novel ethnographic methods developed initially to understand the high rates of errors at home among outpatient children with chronic conditions (Table 1) (41). Ultimately (in subsequent phases of this initiative) findings will inform development and piloting of a communication-based intervention to improve diagnostic safety in this population.

A diagnostic journey as conceptualized in SEIPS 3.0 and the National Academies of Medicine starts at the first symptoms of a new problem at home leading to engagement with the healthcare system (1, 35). The healthcare team then engages in an iterative process of information gathering, information integration/interpretation, and formulation of a working diagnosis which is communicated to the patient and family and revisited as needed in response to treatment; success in this process determines outcomes for both the patient and system (1).

We will sample children followed at our primary care or complex care clinics (designed to care for patients with multi-specialty involvement, technology dependence, and/or neurologic impairment). *Our preliminary chart review showed that diagnostic journeys for children with multiple chronic conditions lasted from one day to three weeks.* We thus expect to capture the majority of diagnostic errors within six weeks of the initial presenting visit and will perform chart reviews and parent/caregiver phone surveys at that time. We will re-review all charts six months later to characterize the distribution of the duration of the diagnostic journey and assess the sensitivity of the six-week cutoff.

TABLE 1 Study procedures, timing and purpose of each procedure.

Procedure	Timing	Purpose
Observation of urgent visit (n = up to 35)*	Presentation for urgent problem	Evaluate patient-centered communication quality and content and diagnostic uncertainty. Identify interventional opportunities.
Parent interview (n = up to 35)*	Within 2 weeks of observation	Assess patient-centered communication quality and content, diagnostic uncertainty, and discrimination
Clinician interview (n = up to 35)*	Within 2 weeks of observation	Evaluate patient-centered communication quality and content and diagnostic uncertainty
Chart review (n = 150)	Within 4 weeks, and by 6 months after the last visit for presenting problem	Describe the diagnostic journey. Identify diagnostic success and errors applying standardized patient safety and diagnostic evaluation instruments.
Parent phone survey (n = 150)	Within 4 weeks, and by 6 months after the last visit for presenting problem	Identify elements of the diagnostic journey not recorded in the chart. Identify diagnostic success and error.

*Observations and interviews will be performed until thematic saturation is achieved.

3.2 Setting

Data will be collected in five primary care and complex care ambulatory clinics associated with three children's hospitals in the Northeast and Midwest. Clinic patient characteristics vary. Between 40 and 6% of patients identify as Hispanic, 45 to 27% of patients identify as Black, and 69 to 23% of patients identify as white. The majority of patients have public insurance (range across clinics 55 to 85%). Parental preference for a language other than English ranges from 20 to 8%.

3.3 Participant selection

We will recruit 150 patients age < 21 years with multiple chronic conditions (excluding mild asthma, eczema, allergies) presenting with irritability, vomiting, fever, abdominal pain, or other acute illness. We will review records of and conduct a brief phone survey (family safety reporting) with all participants. For the qualitative phase of the study, we will observe up to 35 of these patients' clinic visits and invite

parents/caregivers and a clinician from each observation to participate in interviews and additional, follow up phone surveys. To assure a comprehensive perspective on the diagnostic process, we will only include patients who receive primary care within our health systems. To understand differences in diagnostic processes by patient/family characteristics and parent/caregiver language, we will recruit patients with diverse racial and ethnic backgrounds and oversample patients with caregivers who prefer Spanish. Research assistants from diverse racial, ethnic, and language backgrounds will recruit outpatients by phone or in person. Participants will be compensated for interviews and observations. Consent will be obtained; assent will be obtained from the child where appropriate.

3.4 Methods

Chart reviews and brief phone surveys (150 patients) will be conducted to understand the longitudinal diagnostic journey, identify medical errors/harms, and characterize opportunities to improve diagnosis. Initial reviews will take place within 4 weeks of an acute care visit and will continue, with the goal of characterizing the diagnostic journey in its entirety, for up to 6 months. To further characterize the diagnostic journey, a trained research nurse will abstract the dates and locations of interactions with the health system, including in person, virtual, telephone, and portal message encounters. All pertinent communication and documentation regarding presenting, index-visit symptoms will be abstracted. We will detail "good catches" or "near misses" as sources of resilience. Charts will be reviewed for medical errors, associated harms, and harm severity (2, 42–47). In order to evaluate diagnosis, a structured EHR review tool [Revised SaferDx (48, 49)] will be applied to each case by two clinicians who will determine whether there were missed opportunities to improve diagnosis. Missed opportunities will be further classified using the modified DEER taxonomy to identify the phase of the diagnostic process involved (50). To maximize reliability, reviewers will receive didactic training practice on identical charts (45, 46). We will assess inter-reviewer reliability and discordant reviews will be adjudicated by a third, consensus reviewer.

In light of the inherent limitations of retrospective chart review, we will also conduct brief phone surveys with patients/families to identify aspects of the diagnostic journey not otherwise captured in the medical record including parent/caregiver perceptions of diagnostic excellence, error, communication, and systems resilience. Parent/caregiver phone surveys are routinely used in outpatient safety research to identify care processes and potential errors not recorded in the chart (51, 52). We developed a ten-minute, 15-item phone survey, adapting from previously developed tools to capture family safety reporting in the hospital (4, 5, 53, 54) The survey examines points along the diagnostic journey, including successes, failures, and errors at each point. Surveys will occur within 4 weeks, and by 6 months after the last known point of contact with the health care system for the diagnostic journey (e.g., last outpatient visit, last outpatient follow up after hospitalization, or hospital discharge).

Observations. A subset of up to 35 patients will be recruited to participate in ethnographic observations. Observations will aim to characterize variation in diagnosis-focused communication across the diagnostic process (1) including during the phases of information gathering, information interpretation and integration, and formulation/communication of the working diagnosis (and diagnostic uncertainty, if present). A trained research assistant will be physically present in the exam room, directly observe, and audio-record the patient's entire clinic visit. The Hawthorne effect will be mitigated by having ethnographers shadow providers right before recruitment starts to increase clinic staff's familiarity with the research team, training ethnographers to be inobtrusive, and triangulating data from observations with other data collected from interviews and chart review. We will utilize an iteratively-developed observational guide developed based on conceptual models of patient/family identity, clinician-patient communication and health outcomes (55), Safety II/ resilience engineering (56), and the SEIPS 3.0 framework (35) (Figure 1). Portions of audio-recorded observations will be transcribed at a later date, if needed to understand communication content. Diagnostic successes and errors will be identified within and across SEIPS work system domains (37) and used to model the system and identify resilience (36).

Semi-structured interviews of parents/caregivers and clinicians who participate in observations will be performed by a trained interviewer in person or by phone, within 2 weeks of observation. The goal of these interviews is to reflect on interactions and processes during the observed clinic visit in order to obtain a deep understanding of how communication affects the diagnostic processes in light of patient and family characteristics, circumstances, and values. Interviews will explore parent/caregiver experiences in accessing care, perceived discrimination, and patient-centered communication ("*When did the doctors listen to you?" "not listen*?") throughout the diagnostic process, from history taking to formulation of working diagnosis. Interviews may also ask about adaptive clinician approaches, and variation in parent/caregiver "speaking up." The semi-structured interview guides were developed using the same frameworks as for the observation guide and will be pilot-tested with diverse participants and edited as needed for clarity. Interviews will be conducted by trained bilingual interviewers, audio-recorded, and transcribed by a HIPAA-compliant transcription and translation service. Surveys, administered concurrent to the interviews, will inform interpretation of qualitative data. Parents/caregivers will compete a 3-item health literacy survey (57). Clinicians participating in ethnography will complete the Team Dynamics survey (58).

3.5 Analysis of chart reviews

Descriptive statistics will be used to summarize demographics and aspects of diagnostic journeys, including duration (in days), number and types of contacts within healthcare, number of people, and clinics involved. Rates of medical and diagnostic errors will be estimated. Patient-days will be determined by calculating the number of days between first point of contact with the health system and the most-definitive diagnosis and treatment. Rates will be calculated per patient and per 1,000 patient-days. Tabulation of SaferDx (49) and DEER taxonomy (50) results will be used to ascertain rates and types of diagnostic successes and errors. In addition to establishing a baseline incidence of opportunities to improve diagnosis in this primary care cohort, our analysis will identify areas of vulnerability in the diagnostic process and elucidate the relationship between medical errors, diagnostic errors, and outpatient harm. In conjunction with qualitative analysis, our findings will focus and direct interventional efforts

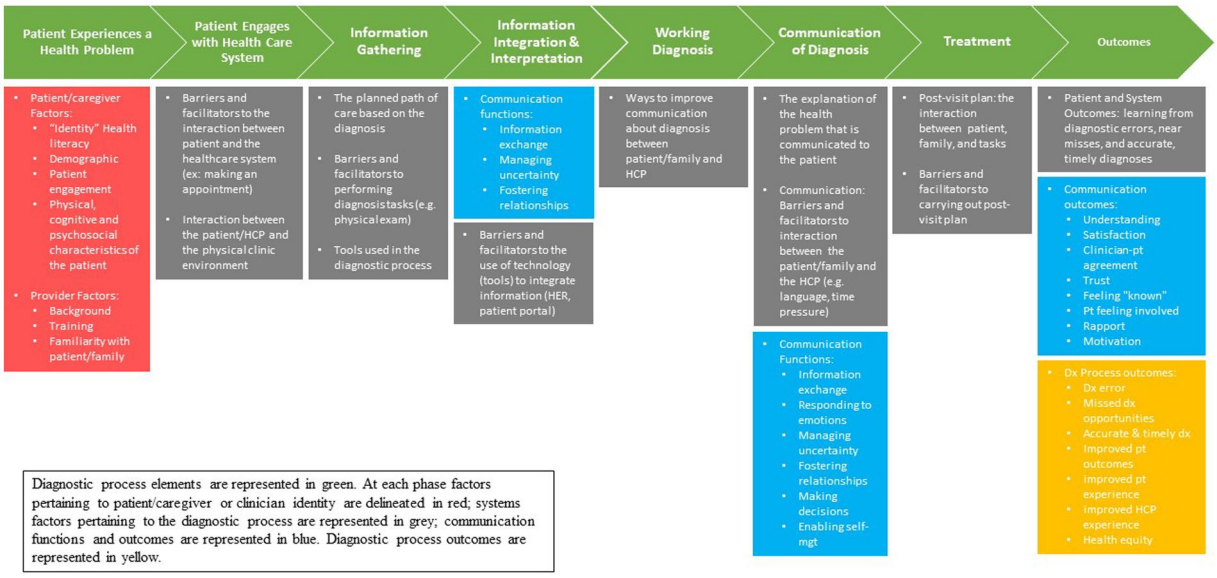

FIGURE 1
Integrated conceptual model of the ways patient/caregiver identity, clinician-patient communication health outcomes, resilience engineering, and the systems engineering for patient safety framework intersect and should be evaluated during the diagnostic process.

targeting opportunities identified in the evaluation of existing diagnostic processes.

Because of the small sample size, we will perform stratified analyses by site only and will not create adjusted models. Given the size of this study and the expected rate of diagnostic errors, we do not expect to quantitatively identify differences in rates of diagnostic errors by parent/caregiver preferred language, insurance status, or self-reported cultural, religious, gender, disabilities, or racial/ethnic identities. However, we do expect to note qualitative differences in diagnostic processes, which will inform future observational and interventional studies. Demographics of participants will be compared with those of eligible non-participants who declined or whom we were unable to consent for participation in order to evaluate for potential selection bias.

3.6 Qualitative analysis of observations and semi-structured interviews

Qualitative analysis will occur in tandem with ongoing observations and interviews to ensure fidelity to the guides, appropriate observation and interviewing techniques, and to enable detection of data saturation (the point at which no new major codes/themes emerge). Analyses will proceed through the development of mixed deductive/inductive codes and coding using the constant comparative method, culminating in overall thematic analysis of the coded data (59). We will develop deductive codes drawing on the literature and conceptual models of patient/family identity clinician-patient communication (55), Safety II (56), and the SEIPS 3.0 framework (35) used to generate the observation guide (Supplemental). Qualitative coding will be performed using Dedoose software (Version 9.0.17, SocioCultural Research Consultants, LLC, Los Angeles, CA, 2021) by assigning deductive (from *a priori* codebook) and inductive codes (from emergent themes) to text segments, grouping related codes, and iteratively revising the coding structure as new codes emerge. Group coding will be performed for qualitative data from the first 2–3 patients, revising the codebook as needed. Thereafter, qualitative data from each patient will be coded independently by two coders, resolving discrepancies at scheduled consensus meetings. We will develop definitions for diagnostic success and errors and use inductive thematic analysis to ascertain systems factors that appear to be contributing to the events. Directed content analysis will be used to identify the ways in which patients, families, and clinicians communicate along the diagnostic journey within and between systems of care. We will note successes, sources of resiliency, errors, and barriers or facilitators of the diagnostic process. We will compare processes and themes along major sub-groups. We will also characterize the diagnostic system, processes, and resilience at each site, comparing the similarities and differences, and thus identifying common themes and specific features.

Our ethnographic and chart review data will inform the development of process maps showing the entire diagnostic journey for children with multiple chronic conditions at each site and across sites, as well as process maps for families using Spanish for care. In our prior studies, such process maps have been pivotal in identifying points for intervention. Moreover, analysis of communication, patient/family-reported opportunities to improve diagnosis, and identification of the sources of resilience that can facilitate diagnostic excellence will focus intervention design on key aspects of diagnostic process.

4 Ethics and dissemination

Findings resulting from the described protocol will provide a basis for the development of interventions and methods to be used broadly in evaluating and improving the provision of diagnostic excellence in pediatric primary care. This study poses minimal risk to participants and has been approved by the single Institutional Review Board at

Boston Children's Hospital upon which the other sites are reliant. Consent and assent will be obtained as described above. In the unlikely event that we identify a serious diagnostic error in evolution or adverse event in evolution, we will address these directly with the clinical team, including the attending physician. The clinical team would follow their usual clinical procedure, including following institution-specific guidelines for error reporting.

5 Discussion

Our methods and outcomes are novel and represent an effort to evaluate and improve diagnostic communication longitudinally across time and care settings. Findings will help hone our understanding of the relationships between pediatric patients with medical complexity and clinicians, the ways patient/caregiver racial, ethnic, and linguistic identities intersect with communication and diagnostic outcomes, and the aspects of care that enable diagnostic excellence. With ethnography we will be able to evaluate systems resilience and when diagnostic communication processes "go well," using a Safety II lens. In doing so, we will extend the idea that the diagnostic process is the product of communications situated in systems-of-work that are constantly adapting to the everyday challenges. Both ethnographic and chart review methods are meant to test and refine tools for identifying opportunities to improve diagnosis (Safety I). More broadly, we will assess the validity of different methods to detect diagnostic errors along the diagnostic journey. An outcome of this work will be evaluation and reflection on our methodology for detecting systems resilience, for detecting opportunities to improve diagnosis in primary care, for achieving across-site concordance in identifying opportunities to improve diagnosis, and for leveraging interdisciplinary expertise to evaluate cases. Our approach will allow us to learn about the diagnostic process for these vulnerable patients in primary care, but also extend and refine methods to be used to evaluate diagnostic performance and equity.

Ethics statement

The studies involving humans have been approved by the Single Institutional Review Board at Boston Children's Hospital. The studies will be conducted in accordance with the local legislation and institutional requirements. The participants' guardians/next of kin will provide their written informed consent to participate prior to the commencement of the study and assent will be obtained from the child where appropriate.

Author contributions

IR: Conceptualization, Funding acquisition, Writing – original draft. TM: Funding acquisition, Methodology, Writing – review & editing. CC: Funding acquisition, Methodology, Writing – review & editing. KC: Methodology, Writing – review & editing. NK: Conceptualization, Writing – review & editing. PB: Funding acquisition, Methodology, Writing – review & editing. KM: Writing – review & editing, Methodology, Funding acquisition. AK: Funding acquisition, Methodology, Writing – review & editing. AC: Writing – review & editing, Funding acquisition, Methodology. EL: Writing – review & editing, Funding acquisition, Methodology. CL: Funding acquisition, Methodology, Writing – review & editing. KW: Funding acquisition, Methodology, Writing – review & editing.

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Conflict of interest

Kuzma is a consultant for the I-PASS Patient Safety Institute. The I-PASS Patient Safety Institute is a company that seeks to train institutions in best handoff practices and aid in their implementation. Landrigan has consulted with and holds equity in the I-PASS Institute, which seeks to train institutions in best handoff practices and aid in their implementation. In addition, Landrigan has received monetary awards, honoraria, and travel reimbursement from multiple academic and professional organizations for teaching and consulting on sleep deprivation, physician performance, handoffs, and safety, and has served as an expert witness in cases regarding patient safety and sleep deprivation. Walsh has served as a consultant for Sanofi and Research Triangle Institute.

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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Framing diagnostic error: an epidemiological perspective

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Diagnostic errors burden the United States healthcare system. Depending on how they are defined, between 40,000 and 4 million cases occur annually. Despite this striking statistic, and the potential benefits epidemiological approaches offer in identifying risk factors for sub-optimal diagnoses, diagnostic error remains an underprioritized epidemiolocal research topic. Magnifying the challenge are the array of forms and definitions of diagnostic errors, and limited sources of data documenting their occurrence. In this narrative review, we outline a framework for improving epidemiological applications in understanding risk factors for diagnostic error. This includes explicitly defining diagnostic error, specifying the hypothesis and research questions, consideration of systemic including social and economic factors, as well as the time-dependency of diagnosis relative to disease progression. Additional considerations for future epidemiological research on diagnostic errors include establishing standardized research databases, as well as identifying potential important sources of study bias.

KEYWORDS

epidemiology, medical misdiagnosis, health equity, diagnostic error, risk factors, societal implications

Introduction

Diagnostic errors remain a grossly under-assessed patient safety and quality of care threat (1–3). Annual estimates of diagnostic error vary widely, from 40,000 to 4 million cases nationwide. Over half are related to cardiovascular diseases, infections, and cancers, and over 6% of these result in serious harm to patients (4). Newman-Toker et al. estimated that 5.7% of all emergency department visits in the United States involve some diagnostic error, affecting 7 million patients annually. Furthermore, 0.3% of all patients suffer from related preventable and serious harm, including disability or death (5). Notably, the published literature tends to focus on medication errors, surgical complications, or healthcare-acquired infections rather than medical diagnostic error (6).

Challenges in evaluating the etiological underlying causes of diagnostic error stem from methodological, logistical and social considerations. Factors such as the emergence of new diseases, evolution of diagnostic capabilities, and advancement of clinical medicine and research complicate defining diagnostic error (7). Initial disease diagnosis may be more subjective due to a lack of confirmatory testing, and greater reliance on provider experience and medical cognition. Diagnostic errors can be obscured when multiple clinicians concur with a given diagnosis and sometimes only revealed retrospectively or after a complaint is raised (3, 8). Moreover, variations in clinical thresholds for testing (including resource limitations) undermine standardization. Consequently, definitions of diagnostic errors may

be subject to the perspectives and responsibilities of the defining stakeholder; clinicians, patients, and researchers likely have different measures and purposes for defining events (or omissions) as "errors."

Epidemiological analysis can reveal patterns in rates, types, risk factors and root causes. A standard framework using epidemiological tools can be applied across diseases and clinical presentations and settings however, all such research will require valid definitions of the diagnostic error outcomes. Ultimately, identifying the factors that result in diagnostic errors subsequently can inform strategic mitigation approaches and improve patient safety.

In this narrative review, we present several dimensions for improving epidemiological research on diagnostic error. Key considerations include accuracy of diagnosis, the relative timing of diagnosis, data sources and their accuracy and completeness, social determinants of diagnostic options, and potential sources of bias including those associated with the patient (e.g., obesity, race, affluence, etc.), the provider (e.g., training and experience, level of interest and commitment, etc.) and the clinical setting (e.g., academic vs. commercial institutions, business and revenue models, insurance structures, etc.). Our primary objective is to illuminate the complex landscape of diagnostic error research, highlighting the nuanced challenges of using epidemiologic methods to analyze the roots of diagnostic error.

Defining diagnostic error

The Institute of Medicine (IOM) defines diagnostic error as "*the failure to (a) establish an accurate and timely explanation of the patient's health problem(s) or (b) communicate that explanation to the patient*" (9, 10). However, this is one of several definitions (see Table 1). Schiff et al. (2009) offers greater scope, defining diagnostic error as "any mistake or failure in the diagnostic process leading to a misdiagnosis, a missed diagnosis, or a delayed diagnosis (11)." This broader definition highlights diverse types of errors, such as wrong, overlooked and delayed diagnosis, and considers both the method and timing of diagnosis (11) (Table 1).

The diagnostic process is complex, evolving over time and involving multiple stakeholders, which can complicate finding the sources of error (12). This complexity introduces an intriguing challenge: addressing diagnostic error as a time-dependent phenomenon. Therefore, "error" can be defined in two ways: deviation from "the truth" or, departure from what reasonably (or expertly) could be achieved based on the available information at specific stages of the disease. For instance, at the earliest stages of a disease process, diagnostic ability is limited by the available indications, but as the disease progresses, additional test results, signs and symptoms may provide increasingly clear diagnostic clues.

A standardized definition of diagnostic error is important for both comparability across studies and validity and reproducibility of individual study findings.

TABLE 1 Diagnostic error: select definitions (1, 9–11, 19, 32–34).

Term	Definition	Citation
Diagnostic error	"The failure to (a) establish an accurate and timely explanation of the patient's health problem(s) or (b) communicate that explanation to the patient"	IOM
Misdiagnosis-related harms	"Harms resulting from the delay or failure to treat a condition actually present (false-negative diagnosis) or from treatment provided for a condition not actually present (false-positive diagnosis)"	Newman-Toker et al. (2009) and (2014)
Diagnostic error	"Diagnosis that was unintentionally delayed (sufficient information was available earlier), wrong (another diagnosis was made before the correct one), or missed (no diagnosis was ever made), as judged from the eventual appreciation of more definitive information"	Graber et al. (2005)
Diagnostic error	"Any mistake or failure in the diagnostic process leading to a misdiagnosis, a missed diagnosis, or a delayed diagnosis"	Schiff et al. (2009)
Diagnostic error	"Implies that something different could have been done to make the correct diagnosis earlier. Evidence of omission (failure to do the right thing) or commission (doing something wrong) exists at the particular point in time at which the 'error' occurred"	Shojania et al. (2003)
Diagnostic error	"1. Case analysis reveals evidence of a missed opportunity to make a correct or timely diagnosis. The concept of a missed opportunity implies that something different could have been done to make the correct diagnosis earlier. The missed opportunity may result from cognitive and/or system factors or may be attributable to more blatant factors, such as lapses in accountability or clear evidence of liability or negligence. 2. Missed opportunity is framed within the context of an "evolving" diagnostic process. The determination of error depends on the temporal or sequential context of events. Evidence of omission (failure to do the right thing) or commission (doing something wrong) exists at the particular point in time at which the "error" occurred. 3. The opportunity could be missed by the provider, care team, system, and/or patient. A preventable error or delay in diagnosis may occur because of factors outside the clinician's immediate control or when a clinician's performance is not contributory. This criterion suggests a system-centric versus physician-centric approach to diagnostic error."	Singh et al. (2005)
Diagnostic error	Undesirable diagnostic events as specific, measurable, and actionable clinical situations likely to denote the presence of diagnostic error.	Olsen et al. (2018)

Partitioning diagnostic error

Considering the range of possible sources of diagnostic error may help refine the hypotheses and research questions in designing epidemiological studies of diagnostic error. Graber et al. (2005) further classified diagnostic errors into three categories: no-fault errors, system-related errors, and cognitive errors (8).

No-fault errors, as described by Kassirer and Kopelman, are influenced by factors that are outside of clinician or system control, such as atypical disease presentation and patient-related factors. These include inaccurate or incomplete information that leads clinicians down an incorrect diagnostic reasoning path (13). Factors related to the patient, healthcare team, or care environment may also compromise diagnostic acuity.

System-related errors are those caused by technical failures, equipment problems, organizational challenges, lack of communication among healthcare teams, or inefficient processes at the system-level (1, 7). The harms of missed diagnoses or misdiagnoses from system-related errors may be more pronounced among marginalized populations, such as racial minorities or the older adults, due to implicit biases or assumptions that impede clinical reasoning.

Cognitive errors are clinician-based mistakes made despite the clinician possessing the ability to make the correct diagnosis, possibly due to subconscious biases (14). They are rooted in implicit biases, confirmation biases, inadequate training or knowledge, poor critical thinking or competency, or failure to fully investigate and gather information (1, 13). The bias of anchoring to premature or initial findings or recent and memorable interactions can create a confirmation bias favoring the evidence of a working hypothesis the clinician may have already had. Additionally, fatigue, stress, or burnout may reduce diagnostic capacity (6).

Diagnostic error likely occurs as the product of multiple partitioning factors occurring simultaneously, including individual perspectives, patient and disease complexities, and systematic practices. Epidemiological study designs and analytical approaches will need to anticipate and accommodate not only easily measured medical and patient characteristics, but also more abstract behavioral, cultural and likely interdependent (i.e., non-independent) variables and factors.

Epidemiological framework

In traditional etiological epidemiological research, the goal often is to identify factors associated with a disease outcome that, if removed, would have prevented that outcome from occurring. Similarly, one might frame this in terms of how the diagnostic outcome would have differed had one or more modifiable "risk" factors not operated or been present. However, diagnostic errors present an additional challenge: they may never be detected or may be impossible to measure directly. This makes it particularly difficult to establish a reliable referent or "gold standard"—also counterfactual—against which the observed diagnostic is compared. Where a disease or disease process is evolving, the expectation that a firm and accurate diagnosis is uniformly possible may be unrealistic. Furthermore, diagnosis is fluid, and often is reformulated, revised or refined over time; therefore ultimately, the diagnostic error *per se* may not solely represent the presumably adverse outcome, but rather, the consequences (possibly in terms of actions or inaction) that result from it. This expands the scope of "risk" factors for diagnostic error to include information that might not be available or obtainable in a given situation (for any number of reasons): in theory, had it been available a more favorable outcome might have been possible.

Therefore, rather than striving for a standardized definition of diagnostic error, we recommend critically thinking about how timing and accuracy of a diagnosis impact its accuracy and validity, and further, what the consequences would be of treatment decisions that rely on the current diagnosis; the possible associated partitioning errors; the validity of available (or derived) data sources and their utility for identifying and characterizing errors; potential sources of biases; and key sociodemographic aspects that might play a role. This provides a general critical framework for studying diagnostic error with epidemiological methods, but with special consideration of the 'non-traditional' study aspects, as well as the flexibility required to address complex systems in which diagnoses are generated, and the diverse responses they may impact. Well known in epidemiology is the potential harm associated with early screening and detection of diseases for which early treatment is not measurably better, especially if psychological stress is induced. Timing and accuracy could then be considered potential confounders or effect modifiers, respectively.

Under the suggested framework, patient-provider interactions that delay the communication of a diagnosis to the patient should be analyzed as if the timing of diagnosis is when the patient received the diagnosis. In situations where the initial diagnosis is revised or improved, additional time to accurate diagnosis accrues. The timing of communication of diagnosis to the patient is important because it marks the beginning of a patient's ability to self-manage their condition, which can affect their ultimate health outcome.

As in all epidemiological applications, study design will vary based on the proposed research questions and objectives. If the primary aim is to understand the overall impact of diagnostic error on patient outcomes, focusing on accuracy might be more appropriate, irrespective of timing, as well as how the diagnosis influenced treatment or other care. Alternatively, if evaluating the timing of diagnostic error is the objective, a baseline timeline or scale will need to be derived (as a referent), such that diagnoses rendered at various times over the natural history of a disease process appropriately will be compared with what might be attainable at each stage. Consequently, the point at which a patient seeks medical care may affect the diagnostic process and the likelihood of diagnostic error. Seeking care may be delayed until symptoms are pronounced, which might make the diagnosis more straightforward. On the other hand, more advanced disease may be accompanied by comorbidities, or reflect atypical signs and symptoms, and in general, over time, underlying risk factors or even causal factors will become distanced from their effects, possibly obscuring the ability to observe a relationship.

Figure 1 presents a simplified directed acyclic graph, or DAG, to illustrate the hypothetical causal relationships between timing or accuracy and diagnostic error. Other nodes may need to be added based on the population of interest. This diagram should be used in conjunction with a clearly defined study design, appropriate statistical analysis, and explicitly stated assumptions

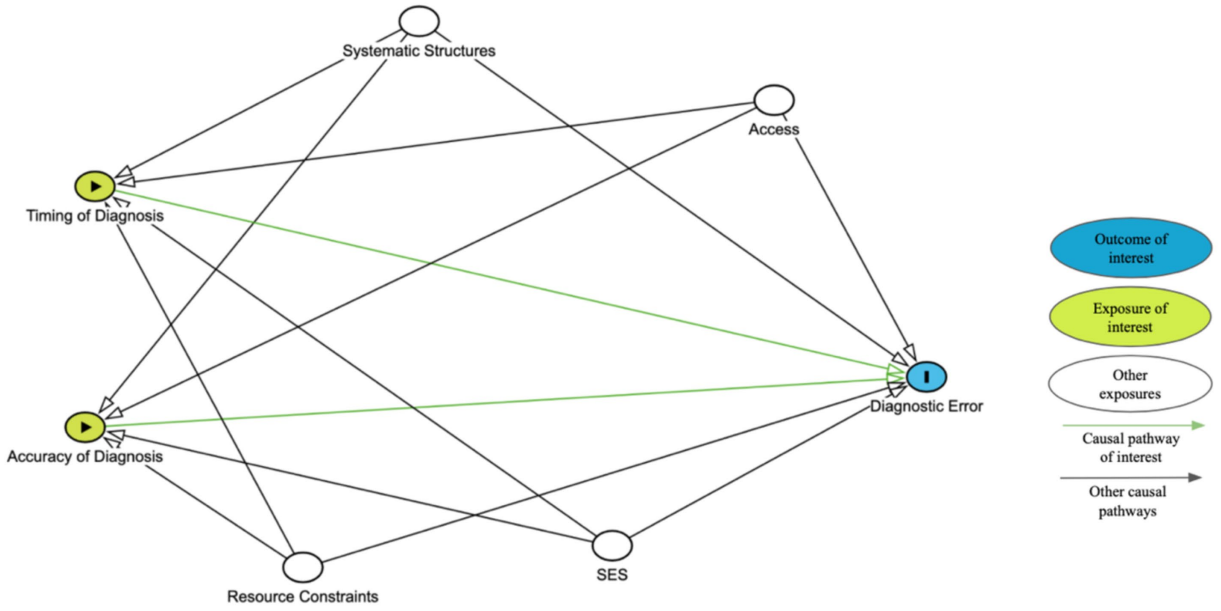

FIGURE 1
Simplified DAG template to guide analyses.

to determine the strength of association between timing or accuracy of diagnosis and diagnostic error.

We acknowledge that the relationship between timing and diagnostic acuity is complex making it difficult to separate the effects. Advanced epidemiological analytical techniques such as path analysis may help researchers clarify the direct and indirect effects of timing and accuracy on diagnostic errors and patient outcomes. However, the decision should be guided by the specific research questions, available data, and understanding of these and other possibly intercorrelated factors. Maintaining transparency in the study methods allows for interpretable, replicable, and ideally, more applicable and improved results.

Other framework considerations

Data sources

Studying diagnostic error from an epidemiological perspective necessitates careful evaluation of available data sources, each with their possible strengths and limitations. These sources could include electronic health records, laboratory and imaging tests, autopsy reports, and health insurance or malpractice claims data—or more subjective sources such as patient care feedback or research-initiated patient surveys. By understanding the specific contexts in which misdiagnoses commonly occur, it is hoped that healthcare systems will be better equipped to devise targeted strategies to prevent these errors (15, 16).

The largest data sources for examining diagnostic errors are administrative databases such as EPIC or Cerner (17). These repositories of medical information are collected and maintained by hospitals, clinics, and insurance companies that include information such as medical claims, records of service, prescriptions, procedures, and diagnoses of patient's medical encounter (14). Retrospective analysis of these data allows researchers and clinicians to assess geographic, demographic, and temporal patterns of diagnostic error. It also facilitates the exploration of system-level factors, such as the relationship between medical institutions' staffing or patient volume and the rate of diagnostic error. Using large administrative databases also allows estimation of population-level diagnostic error rates across medical conditions, the investigation of sociodemographic disparities in diagnostic error, and the study of diagnostic error's impact on care progression. However, there are challenges to the retrospective use of administrative databases. These include the time-consuming nature of review, as sometimes manual review is required for paper-based documentation, and the potential for incomplete or inaccurate information, which may affect the sensitivity and specificity of the method. These limitations can restrict the epidemiological methods available for data analysis (17).

Autopsy reports, pathology findings, clinical health summaries, and interpretative summaries, often reveal significant diagnostic errors (18). It was estimated that historically, as much as 25% of autopsies uncover some form of major diagnostic error, and 10% revealed errors directly contributing to the death; encouragingly, the rate of diagnostic errors discovered at autopsy has declined (19). Schwanda-Burger et al., assessed diagnostic errors in a teaching hospital and reported a 23% decrease in errors found in autopsies over a 30-year period, attributing this decline to improved technology, medical records, and diagnostic tools (20).

Much like large administrative databases, medical malpractice claims data can provide insight into the geographic, demographic, and temporal context of misdiagnoses. These data also can reveal which conditions are most frequently misdiagnosed, although they likely reflect selection biases favoring documented examples where serious harm resulted. For instance, vascular events, infections, and cancers

constituted 72% of all emergency department diagnostic errors resulting in serious harm (5). Patients with more serious conditions or harmful outcomes are more likely to file malpractice claims, whereas those who did not experience serious harm are less likely to file claims (9). However, medical malpractice claims data may exclude groups who lack the knowledge or financial ability to file malpractice claims. Such claims also might be falsely brought, motivated by potential monetary gains. There may also be gaps related to the misdiagnosed condition and the diagnostic error's outcome.

When designing studies and interpreting results, epidemiologists carefully should consider the strengths and weaknesses of available data resources. Understanding the completeness, inherent biases, uncertainties and ultimately the validity of selected data sources can lead to the improvement of study methods and the interpretation of findings. By being transparent, epidemiologists can contribute to a better understanding not only of factors associated with diagnostic error, but to recommendations on improving the approaches and study designs used to evaluate diagnostic errors and their risk factors and possibly their root causes.

Social determinants

Epidemiological research to identify risk factors for diagnostic error additionally requires definition and measurement of a range of indicators and phenomena including individual patient constitutional characteristics (e.g., age, sex, race/ethnicity, educational level, economic access, etc.), location characteristics (e.g., urban, rural, state/region, country); clinical factors (e.g., type of facility, provider attributes including occupation, specialty, education, etc.); access and other socioeconomic factors; and diseases or conditions of interest that may not be well-known or defined. Though there are several social determinants to consider, we note that access to care, resource constraints, and systemic structures can significantly impact timing and accuracy of diagnosis and diagnostic error.

Access

Access to medical care impacts patients' ability to be diagnosed and treated, and likely the quality of these as well. The ability to access and obtain medical care from competent and resourced local providers is not universal, even in highly advanced societies. Medical costs, insurance, transportation, mobility, desire and fear are just a few of the factors that may influence an individual's access to medical attention that could benefit their present and future well-being (21). Greater access to care allows an individual to see a provider sooner and possibly to see multiple providers, theoretically improving the probability of obtaining accurate diagnoses and treatments, including seeking second opinions. Thus, access to medical care should be measured and explored as potential risk factors or effect modifiers in epidemiological studies of diagnostic error.

Resource constraints

Medical care quality may be constrained by institutional resource availability and management. High-and low-resource settings, regions, or systems where healthcare resources are abundant or limited, respectively, are accessed by an institution's financial capacity, technology, workforce ability, care quality, and research program. A region or institution with access to more testing, information, and colleagues to discuss complex patient presentations, allows (at least conceptually) for more accurate and timely diagnoses (22). When the resource setting has more diverse and culturally competent healthcare workers, cultural and language barriers can be more appropriately addressed. This allows patients to feel more capable of effectively participating in their care because the diagnostician is more likely to listen to and understand the patient's symptoms, personal situation, sociodemographic background, cultural perspectives, and beliefs that inform the case and associated research, which is key for better diagnostic care (22). Nevertheless, for more common and less serious medical conditions, no more than basic clinical resources and personnel may be required for accurate diagnoses and appropriate treatment; therefore, the level of consideration of resource constraints also will depend on the research questions epidemiologically being addressed.

Systemic structures

Systemic structures of healthcare involve the interplay of people, resources, processes and institutions, including critical communication pathways among these and ultimately with the patient (23). In epidemiology, these systemic structures are often considered by controlling for confounders such as insurance type or hospital location, tangible entities that are measurable. However, systemic structures are often intangible like power dynamics and agendas of larger institutions that are complex and challenging to define (24). These systemic structures influence the way diagnostic processes are designed, structured, and evaluated. Systemic structures influence the allocation of resources, funding, and training which can impact the accuracy and timing of diagnoses (25). When establishing a study design, it is crucial to acknowledge and account for social determinants (e.g., socioeconomic status, race) that act as proxies for systemic structures. Explicitly identifying how the social determinant(s) might influence diagnostic accuracy or error can help to elucidate these complicated relationships and reveal that certain factors require more complex adjustment.

Potential study biases

Bias in epidemiological research refers to methodological or systematic errors that lead to a distortion in the estimated numerical study result, most often a form of relative risk. Biases can lead to errors that range from inconsequential to profound, i.e., where the result misleadingly indicates a risk where one might not exist—or vice versa. Therefore, the design, planning and execution of an epidemiological study provides opportunities to anticipate and reduce the occurrence—or the impact of—specific potential sources of bias. Accordingly, potential sources of bias in evaluating associations between risk factors and the diagnostic error outcomes can be anticipated, identified and mitigated to increase study validity. For example, as discussed under data sources, legal claims databases likely represent a biased subset of medical misdiagnoses, i.e., those more

likely to have been well documented, those resulting in serious harm, those occurring among litigious groups and those with the legal resources to pursue their cases. It is likely that this group is select and is not representative of the universe of comparable diagnostic errors.

Medical malpractice is a major concern of healthcare practitioners and has been associated with "defensive medicine" including excessive laboratory testing and increased referrals (16). Over-testing and (over) diagnosis can make identifying and assessing the potential role of risk, confounding and effect modifying (i.e., interdependent) factors challenging. Additionally, clinicians may be more cautious when diagnosing patients with certain characteristics or risk factors, introducing diagnostic bias. Ensuring there are non-punitive reporting systems in place to create and promote safety also would reduce bias. A highlight of this system is maintaining elements of anonymity and confidentiality that can help reduce fear of individual provider blame and encourage more accurate reporting of diagnostic errors. A shift of focus to system-level factors, rather than individual-provider-level factors, may also allow for identification of broader issues that affect diagnostic accuracy.

Nevertheless, The World Health Organization (WHO) identified the training of healthcare providers as a contributor to diagnostic error (26). WHO emphasizes that suboptimal training, specifically lack of training for clinical reasoning and deficient certification and licensure training, contributes to diagnostic error and suggests that clinical training such as embedding decision support tools to assist with differential diagnoses could improve reliable diagnosis. Additionally, training in clinical reasoning, patient safety, critical thinking, and cognitive heuristics may also improve diagnostic accuracy (22). It should be highlighted that no-fault errors such as hours worked, fatigue, management style, and compensation also can impact clinician errors (27); however, it is unclear how these factors would introduce bias into epidemiological studies and whether they might be viewed as independent risk factors or effect modifiers.

Previously discussed as a partitioning effect, cognitive biases further can be categorized as availability heuristic, anchoring heuristic, framing effects, and blind obedience, to name a few (28). Availability heuristic results in the diagnosis of a patient based on provider experience with past cases regardless of the patient's current presentation. In a randomized control trial of 46 resident physicians, authors found that physicians who were preemptively presented information about dengue fever were more likely to misdiagnose than physicians that had no previous information presented to them (29).

Anchoring heuristic, the reliance on initial diagnostic impressions, despite subsequent information to the contrary, can stem from overconfidence, lower tolerance to risk, and information availability, which is associated with increased diagnostic error. Healthcare workers can unconsciously note data or impressions that "fit" a given diagnosis, but other clues might be discounted (30).

The framing effect is a diagnostic decision due to subtle cues and collateral information. For example, a patient with a history of opioid use and abdominal pain might be treated for opioid withdrawal when they really had a perforation of the bowels (30).

Finally, blind obedience, i.e., placing undue reliance on a test of opinion of "expert" (31). In a behavioral study of paramedic and respiratory therapy students, it was intentionally necessary for students to challenge authority to prevent patient harm. Authors reported that displacement of responsibility was most influential for some participants in not challenging the authority figure's decision (30). Cognitive biases are common and can significantly influence the diagnostic process leading to patient harm and skew epidemiological data by acting as confounding factors, impacting generalizability, and challenging reproducibility of the study. To minimize the impact of cognitive biases in epidemiological studies, researchers should identify the potential biases in their study design and analysis and understand the role that they may play, addressing the bias accordingly. Alternatively, stratification or more complex analytical parameters such as interaction terms may be needed.

Discussion

In traditional epidemiology, we often design studies that primarily consider one (or very few) exposure(s) or one (or very few) outcome(s)—often classified binarily, e.g., hepatocellular cancer (yes/no) and hepatitis A/B infections, and several potential confounding factors (e.g., alcohol consumption, cigarette smoking, etc.). Once the specific outcome, risk factor(s) and potential confounding factors are identified, they can be measured and the relationships among them explored and summarized. In diagnostic epidemiology, the definition of the outcome needs to be comparably specific; however, as we discussed above, the definition of diagnostic errors likely will need to consider time-and context-specific aspects—in other words, the diagnostic error may not simply be described accurately in absolute or binary terms of "yes/no." Additionally, the risk factors and underlying causes of diagnostic error (in general, and also likely for very specific sub-types) will not be measurable chemical or biological agents, but rather multiple abstract and intercorrelated factors including behavioral, social and systemic influences It will be crucial to define these objectively and measure them accurately in diagnostic epidemiological studies.

The timing of diagnosis is especially important as it significantly influences the diagnostic process and most importantly, downstream clinical actions and outcomes. A delayed diagnosis may reduce the risk of an early incorrect one, but also can allow the condition to worsen, leading to complications or atypical presentations that obscure the ultimate diagnosis. Researchers must carefully define what constitutes a "late" diagnosis for their study context. Conversely, accuracy reflects how well the provider's diagnosis matches the patient's true underlying condition given the available information at that time. This likely will be challenging to ascertain retrospectively, and therefore might require real-time assessment. Both timing and accuracy themselves may be influenced by several identifiable and measurable factors that impact the ultimate diagnosis. Furthermore, the likely contributors to and determinants of diagnostic errors may act independently—in which case they in theory can be measured and statistically controlled—or effect modifiers, which requires more complex statistical treatment.

Despite the inherent challenges, employing epidemiological principles and methods provides a path forward for improved understanding of diagnostic error from a group (vs. individual patient) level, with promising applications to identifying risk factors and improving patient safety. Ultimately, clearly articulating the research question, defining the specific "diagnostic errors" being evaluated, and leveraging appropriate statistical analytical methods will generate more reproducible research that provides

insight into the causes of and factors associated with diagnostic error. The epidemiological approach and methods offer a powerful framework for improving diagnostic science and ultimately healthcare quality.

Data availability statement

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

Author contributions

MH: Conceptualization, Resources, Visualization, Writing – original draft, Writing – review & editing. CS: Writing – original draft, Writing – review & editing, Conceptualization, Resources, Visualization. KM: Supervision, Writing – review & editing, 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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Exploring sociodemographic disparities in diagnostic problems and mistakes in the quest for diagnostic equity: insights from a national survey of patient experiences

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Introduction: As part of building a platform for epidemiological research on diagnostic errors and problems that centers on patients and equity, this paper summarizes the development and analysis of data collected from fielding a survey in a nationally representative U.S. population to explore the prevalence and harm consequences of diagnostic problems or mistakes (referred to here as "diagnostic P&Ms") by respondent-reported sociodemographic characteristics.

Methods: We applied narrative elicitation methods to enhance the rigor of implementing a novel survey about diagnostic experiences. We conducted a U.S. population-based survey of a nationally representative sample in 2022–2023, drawn from the NORC AmeriSpeak® panel. We conducted multivariate regression analysis at the household level and in a patient subsample to explore sociodemographic predictors of diagnostic P&Ms and related outcomes in the aftermath.

Results: The comparative analysis by sociodemographic characteristics estimates prevalence of diagnostic P&Ms, prevalence of persisting harms, rate of respondent-reported perceptions of personal attribute adversely affecting diagnosis, and concern about future diagnostic P&Ms. Outcome estimates ranged from about 4% (concern about future diagnostic P&M) to 38% (at least one P&M in households during the past 4 years). Several sociodemographic groups experienced statistically significant higher levels of risk for these outcomes, with some at greater than twice the odds compared to reference groups—transgender and gender independent individuals (e.g., 5 + –fold odds of expectation of future P&M compared to cis-males), cis-females (e.g., greater than 1.5 odds of persistent physical and emotional harms compared to cis-males), low household income (e.g., twice the likelihood of multiple P&Ms for incomes under $60 K compared to $100 K+ households), younger age (3-fold odds of at least one diagnostic P&M for those under 25 years old compared to those aged 45–54), multiracial individuals (about twice the odds of diagnostic P&Ms compared to non-Hispanic White), and disabled and unable to work full-

time (more than twice the likelihood of perceiving that a personal attribute impaired diagnosis compared to those with other work status designations).

Discussion: This new survey and accompanying data source facilitate an enriched exploration of the patterns of diagnostic disparities and points of leverage through which diagnostic experiences can be made more equitable.

KEYWORDS

patient experience, diagnostic equity, population-based survey, patient safety, diagnostic errors, household survey methodology, sociodemographic risk factors

1 Introduction

Diagnostic errors pose significant risks to public health, contributing to adverse patient outcomes and systemic inefficiencies (1). Despite growing recognition of their impact (2, 3), and new studies documenting the aggregate scope of diagnostic errors in the United States (4), there remains a notable paucity of studies quantifying how the risk of diagnostic errors varies among different population subgroups (5). To date, the research demonstrating the heightened vulnerability for some populations is largely based on data from particular practice sites, practice settings, or health conditions (6–8). Because methods are inconsistent across these studies, the evidence on diagnostic inequities remains fragmented and inadequate in several ways.

First and foremost, most extant studies identifying diagnostic inequities do so using clinical markers, rather than patients' own assessments. Because key aspects of diagnosis—including the effectiveness of clinicians' communication and their responsiveness to patient-reported symptoms, concerns, and experiences—can *only* be reliably assessed through patients' reports, many potential manifestations of diagnostic inequity remain underexamined (5, 9, 10). Moreover, when patients perceive that some personal attribute has deleteriously affected their diagnosis, the potential damage to their relationships with clinicians and/or the wider health care system can lead to persisting loss of trust in medical care, increased concern about misdiagnosis in the future, and weakening of the therapeutic alliance between patients and clinicians essential for accurate and timely diagnosis (11–13).

Second, the handful of published studies that do include patient-reported diagnostic errors and problems (14–17) have relied on relatively small samples. This makes it difficult to sort out which attributes of combined sociodemographic constructs, such as lower socio-economic status (e.g., limited education and limited income reported together in one variable) represent the real predictors of the identified inequities. Because past studies have identified a number of intercorrelated sociodemographic characteristics such as age, gender and sex, race and ethnicity, disability, and economic status—each of which has been individually associated with elevated diagnostic risks—the intersections and interactions of attributes have not, to date, been effectively parsed out (7, 8, 14, 18).

Finally, because the evidence-base currently documenting diagnostic inequities is aggregated from a set of narrowly focused studies, it has been impossible to reliably compare the magnitude of diagnostic shortfalls or harms across different subgroups. This undermines efforts to prioritize among interventions that might reduce diagnostic inequities, because they cannot be sensibly targeted to the groups experiencing the greatest current burdens.

To address these gaps in our understanding of diagnostic disparities and inequities, we developed a novel survey specifically designed for learning more about patients' and household care partners' assessments of diagnostic experiences. Our survey, developed and fielded in a national panel, provides the first ever household-reported data set for comprehensive analysis of "diagnostic problems and/or mistakes" (abbreviated as "diagnostic P&Ms") in the United States to reflect the lived experience of patients and their household care partners. These "diagnostic P&Ms" refer to any problem and/or mistake identified by patients themselves or the people living with them (hereafter referred to as household care partners). They include diagnostic P&M events that "can be caused by not getting enough information from the patient, not ordering the right tests in a timely way, not reading test results correctly, or doctors not sharing information well enough with one another." (See Exhibit 1.) The broader focus on P&Ms is intended to better align survey responses with elucidating safety threats and informing actions in the quest for diagnostic excellence inclusive of diagnostic equity. Although some diagnostic P&Ms may not equate to clinically adjudicated diagnostic errors, they represent lived experiences with problems and mistakes in the public's experience of diagnosis that can undergo epidemiologic analysis. With a large sample –almost 4,000 households screened to identify 1,500+ events reported as P&Ms related to diagnosis—we can better distinguish among correlated attributes associated with elevated risks or harm.

The analysis presented here assesses the sociodemographic correlates of diagnostic P&Ms at both the household and patient level. In this paper, we present the methodology used to conduct our survey, describe the characteristics of the study population, and analyze the sociodemographic predictors of diagnostic P&Ms along with their subsequent effects on patients. We report on the prevalence of diagnostic P&Ms and persisting harms by sociodemographic factors such as household income, gender identity, age, marital status, education, race and ethnicity, disability work status and urban/rural residence. We also estimate how these same personal attributes are related to respondents' expectations regarding future diagnostic risks.

Understanding the distribution of diagnostic disparities is crucial to inform development of targeted interventions to reduce diagnostic P&Ms and persisting harms, to surface deficiencies in diagnostic excellence (19), and ultimately to improve healthcare outcomes most equitably (5, 20, 21). Furthermore, we discuss the implications of our findings for healthcare practice, health delivery systems, policy, and directions for future research. Through our analysis, we aim to contribute valuable insights to the complex epidemiology of patient-reported diagnostic P&Ms and advance

Outcome Measure	Question Wording
Any Diagnostic Mistake or Problem	Some people find, when they are not feeling well or have a health concern, that it can be a challenge for their doctors or other clinicians to work with them to figure out what health problem they have. It can also be challenging to clearly communicate what that problem is and what should be done next. We will refer to these situations as "diagnostic mistakes or problems". These can lead to delays in diagnosis, diagnoses that are missed entirely, or diagnoses that are not explained well enough for patients and their families to understand them. Diagnostic mistakes or problems can be caused by not getting enough information from the patient, not ordering the right tests in a timely way, not reading test results correctly, or doctors not sharing information well enough with one another. In the past four years, have you or a member of your household experienced any diagnostic mistakes or problems?
Multiple Events Involving Diagnostic Mistakes or Problems	Did more than one diagnostic mistake or problem happen to you/them in the past four years?
Diagnostic Mistake or Problem with Persisting Physical Harms *(Harms for the Patient)*	When the diagnostic mistakes or problems were happening, how was your/their physical health affected overall? Did your/their physical health stay the same, get somewhat worse, get much worse or [in reporting on someone else's diagnosis] did they die? [If somewhat or much worse] Is your/their health still be impacted?
Diagnostic Mistake or Problem with Persisting Emotional Harms *(Harms for the Respondent)*	Now, thinking about the emotional impact of the diagnostic mistake or problem, did you experience anxiety in the months during or immediately after your discovering that there was some problem with the diagnosis? [If yes] Are you still experiencing anxiety?
Diagnostic Mistake or Problem Involving Negative Personal Attributions	Sometimes aspects of people's background, culture, identity or health needs make their diagnostic experiences better or worse. How, if at all, do you think these factors impacted your/their diagnostic experience? [If yes, those who reported at least one negative impact]
Diagnostic Mistake or Problem Very Likely in Future	How likely do you think it is that a diagnostic mistake or problem would occur when you receive health care in the future? Would you say it is… [Those reporting "very likely"]

EXHIBIT 1
Survey question wording for outcome variables.

efforts towards promoting data-informed and patient-centric diagnostic equity.

2 Methods

The survey that generated the data for the analysis presented below was developed to provide a more robust and patient-centric representation of the diagnostic experiences of the American public. Its conceptual foundation closely accords with the one recently published by Bell and colleagues (10), though our approach (a) operationalizes an alternative way of labeling the sorts of experiences that "count" as diagnostic P&Ms, (b) embodies a commitment to rigorously eliciting narrative accounts about those diagnostic experiences, and (c) incorporates attention to patient experiences, outcomes, and expectations in the aftermath of the diagnostic P&M. We describe below the survey development process and the specific wording of key questions.

2.1 Source of the data

Survey data were collected from a randomly selected subset of people participating in NORC's AmeriSpeak® online panel of over 50,000 households, designed to elicit participation from historically underrepresented populations to ensure that respondents are representative of the American public (22, 23). The panel methods used, similar to other online panels, are transparently documented

and frequently assessed for reliability and representativeness (24–27). Online surveying options include two response modes: Computer-Assisted Web Interviewing (CAWI) and Computer-Assisted Telephone Interviewing (CATI). NORC collects and regularly updates information on all panelists, which makes it possible to assess the sociodemographic characteristics of respondents who screened out of the full survey because no one in their household had experienced a diagnostic P&M.

Participants in the AmeriSpeak® panel receive participation points for responding to surveys. Those participating in a survey of the length of the NEP-DE study receive compensation worth approximately $5.00.

2.2 Survey development

The survey used in this study was developed using a three-stage iterative process that began in April 2022 and ran through May 2023. The initial version of the survey built upon prior work on other patient safety concerns (e.g., treatment and medication errors), literature reviews, and team members' extensive experience in survey methods and diagnostic care assessment (17, 28–31). We incorporated a rigorous narrative elicitation protocol (NEP) methodology to construct the question sequence including 10 open-ended questions that encourage a robust, balanced, and complete account from respondents (32–34, 75). It centered an inclusive understanding of patients' and care partners' lived experiences with the diagnostic process and outcomes, not linked to any particular care setting. The survey went through multiple phases of pilot testing, triangulation with qualitative interview data on a subset of respondents, and revisions to establish a robust and feasible set of survey questions. The novel survey is referred to as NEP-DE (see Appendix). This process was supported by input from an advisory group of patient advocates with lived experience with diagnostic P&Ms, clinicians with expertise in diagnosis and in identifying diagnostic P&Ms, and researchers with expertise in the elicitation and assessment of patient narratives.

2.3 Survey questions assessing outcomes

Although some previous patient experience surveys have aspired to identify events that the public views as "diagnostic errors" (35), researchers attentive to patient experience have increasingly recognized that the public views adverse diagnostic events in broader terms (36–38). Indeed, the public does not always relate to the concept of a medical error, even when it is defined for them on a survey (10). To provide a more inclusive scope, our survey asked about experiences with "mistakes and/or problems" during diagnosis (see Exhibit 1). In separate analyses of the survey, we have noted that those diagnostic P&Ms identified by patients or care partners as "problems" have, on average, as frequent and substantial harms as those that they view as "mistakes." In other work, we have also documented that acknowledgement of something going wrong from someone in a healthcare setting occurred in about one out of three P&M reports (39).

To assess the frequency with which diagnostic P&Ms are encountered, we screened respondents aged 18+ from NORC's AmeriSpeak® online panel regarding the diagnostic experiences of people in their households during the previous 4 years. Extended lookback periods are common for surveys of patient experiences involving safety events. Past surveys included lookback periods of 1, 4, 7 and 10 years (16, 35, 38, 40). Our use of a four-year lookback on this survey corresponds to about the midpoint of this range.

In response to the wording in the screening invitation, respondents initially identified whether anyone in their household had experienced a diagnostic P&M in the previous 4 years. Those who responded affirmatively were then asked if there had been more than one such diagnostic P&M during that time period. About half (51%) of those who reported at least one event indicated that there had been multiple events in their household. These were then prioritized by algorithm—diagnostic P&Ms that involved the respondent's own health care were given priority, and respondents were directed to describe the most memorable P&M for themselves. Diagnostic P&Ms in which the respondent had been a household care partner were included only if the respondent had no personal experience about themselves to report. For household care partner reported events, respondents were again guided to select the most memorable P&M to further elaborate what had happened on a single P&M.

Respondents were asked multiple questions about the selected diagnostic safety event. As shown in Exhibit 1, these included whether the event had induced physical harms for the patient that still persisted at the time of the survey, emotional harms for the respondent that still persisted at the time of the survey, and whether the diagnostic experience had been negatively affected by the system or clinicians in relationship to one or more of the patient's personal attributes. This final outcome was quantified on the basis of coding responses to the last open-ended question from the sequence of 10 questions incorporated into the NEP (see Appendix). Finally, patients were asked about their expectations regarding future diagnostic risks; responding on a four-point scale that ranged from "very likely" to "not at all likely" (Exhibit 1).

For illustrative purposes, we assembled excerpts from the open-ended questions (see Appendix) and the responses that related to selected outcomes: diagnostic P&Ms, persisting physical and emotional harms (an indicator of severe impact), and respondent perception that personal attributes impaired diagnosis. Three steps were required to provide examples of each outcome from the two perspectives: patient reports and care partner reports. First, we selected a subset of responses to represent all outcomes of interest. Second, we selected excerpts to reflect a range of writing styles and narrative lengths. Third, we crafted each excerpt using verbatim text with only minor revisions for readability (e.g., capitalizations where appropriate, but no changes to phrasing or words used), and assembling narrative segments for conciseness and continuity without necessarily reflecting the exact order or full text available in the original open-ended response.

2.4 Survey questions assessing sociodemographic predictors

The sociodemographic characteristics used to identify patterns of disparities in diagnostic experiences rely on information collected from all AmeriSpeak® panelists, as well as additional information collected during the survey process (Exhibit 2). Past studies relying on smaller scale or setting-specific samples suggest that certain subgroups

Sociodemographic Variable	Household Sample Source	Patient Sample Source
Patient Age	NEP-DE Survey/ Household care partner asked to report patient's age	NEP-DE Survey/Patient respondent asked to report their own age
Annual Household Income	AmeriSpeak/ self-identified	AmeriSpeak / self-identified
Education Completed	Amerispeak/ self-identified Household care partner response used as proxy for the patient's education completed	AmeriSpeak/ self-identified
Gender	Amerispeak/ self-identified Household care partner's identification of the patient via analysis of pronouns and relational references to the patient from NEP-DE Survey Open- Response Analysis	Amerispeak/ Self-identified
Race & Ethnicity	Amerispeak/self-identified Household care partner response used as proxy for patient's race & ethnicity	Amerispeak/ self-identified
Geographic Location	Amerispeak/ self-identified	Amerispeak/ self-identified
Work Status/ Disabled and Unable to Have Full-Time Work	Amerispeak/ self-identified Household care partner response used as proxy for patient's status	Amerispeak/ self-identified
Marital Status	Amerispeak/ self-identified For household care partner reporting on a spouse, the patient is classified as married. For other household care partner relationships (e.g., parent), the patient is classified as not married	Amerispeak/ self-identified

EXHIBIT 2
Sources for sociodemographic variables.

of respondents are likely to be at heightened risk for diagnostic P&Ms, including patients from ethnic or racial minority groups (8), those with physical disabilities (18), patients from sexual and gender minority groups (41), women (7), younger and older adults (7), and those from disadvantaged socioeconomic households (14). We also include two additional sociodemographic variables that are plausibly related to so-called "upstream" determinants of diagnostic inequities (5, 42): rural residents (who face larger travel burdens in seeking out diagnoses, particularly when these involve specialists) and respondents with more limited educational attainment. Finally, we include marital status as this social factor has been shown to be protective for health outcomes in other contexts (43).

The terminology for sociodemographic categories (Exhibit 2) were chosen to align with NORC's questions and response options (22). In addition, terminology for several subgroups was adapted based on additional sources (44, 45). For example, while we refer to a gender category, we use terminology for three population subcategories to be inclusive of populations who have non-binary gender identities: cis-male, cis-female, and transgender and gender independent (44). This choice aligns with the panelist responses to four choices for the question "how do you describe yourself?"—male, female, transgender or do not identify as male, female or transgender. The multiple races category refers to two or more races, and we use the term multiracial when referring to individuals in this population (45).

2.5 Sample selection

The AmeriSpeak® national panel was utilized to recruit participants for the survey. Panelists were offered the opportunity to complete a "survey about healthcare experiences"; 26.5% of those offered agreed to participate. Out of this participant pool, 43.6% reported having had a household member (oneself or someone else in the household) with some form of diagnostic mistake or problem during the previous 4 years. Of those who screened into the survey based on having a health care experience and then agreeing to participate, 95.4% completed the entire set of questions about adverse experiences with diagnosis.

Because the survey incorporated an extensive set of open-ended questions, which described the nature of the reported diagnostic event, we were able to further screen the reported P&M to ensure that

the problems were in fact associated with diagnosis rather than treatment. Based on analysis of the narrative responses, we excluded 5.6% of the cases reported from the AmeriSpeak® respondents; these can be viewed as "false positives."

The analytic sample was further restricted by two additional considerations. First, because income is reported at the household (shared residence) level, we excluded all P&Ms reported among family members who no longer shared the residence—this excluded a total of 270 reported P&Ms. Second, because the AmeriSpeak® panel includes only respondents 18 and older, we also excluded all cases reported by household care partners that involved patients under the age of 18—this excluded an additional 45 reported P&Ms from this analysis.

2.6 Data collection

We fielded the survey in three waves, the first in April of 2022, the last in May of 2023. After each of the first two waves of data collection, we further refined the question wording and sequencing. The median time to complete the survey was 23 min. Debriefing questions situated at the end of the survey suggested few difficulties in understanding or completing any of the questions.

Changes to the survey included: (a) after the first wave of the survey, altering the way in which respondents identified when they first began to search for a diagnosis in the sentinel case, so that the response included the month as well as the year of initiation, (b) altering the sequencing and/or wording of three of the prompts for the open-ended questions between the second and third waves, and (c) introducing a new question in the second wave which asked respondents to identify whether, at the time they completed the survey, the uncertainties associated with their diagnosis had been fully resolved.

The narrative elicitation sequence on this survey included 10 open-ended questions, six related to the diagnostic process, four to experiences after the respondent had determined that there had been a mistake or problem related to the diagnosis (see Appendix). The mean response time for the open-ended question sequence was 8.5 min, the median 7 min.

2.7 Statistical analysis

We estimated two different sets of regressions. The first set estimated the prevalence of any patient-reported diagnostic P&Ms or persistent harms. Slightly more than half (54.5%) of the cases of diagnostic P&Ms involved respondent-reported care for themselves. The second set of models estimated the prevalence of P&Ms and persistent harms from the full sample for any member of the respondent's household, including reports from both patients and household care partners in the sample.

Multivariate regression models were estimated to identify the relationship between sociodemographic factors and diagnostic outcomes. For binary outcome variables (any P&M, multiple P&Ms, any persisting physical harms, any persisting emotional harms, any evidence that diagnosis was impaired by inappropriate attention to one or more of patient's personal attributes) the regressions were estimated as logistic models. When the perceived future risk of a diagnostic problem was the outcome, the models were estimated as ordered logistic regression.

2.8 Ethical considerations

The study was deemed exempt by the IRBs at Yale (#2000032012) and Johns Hopkins (IRB00322791) universities. The study fell under the umbrella exemption granted by the University of Wisconsin IRB to qualitative projects conducted by the Qualitative and Health Experiences Research Laboratory in the Department of Family Medicine.

2.9 Data availability

Data from this study will be made available upon request from the corresponding author, after June 1, 2025.

3 Results

3.1 Respondent characteristics

A total of 3,995 AmeriSpeak® panelists responded to the screening questions. As shown in Table 1, the sociodemographic characteristics of these respondents mirrored those of the general U.S. adult population as of 2020 with some modest divergences.

The respondents were slightly older with the most marked difference in the 65 and older groups (24.7% versus 21.6% in the general population). Although there were fewer of the youngest adults (18 through 24 years old) (9.8% versus 12.1%), the proportions of those under the age of 35 years old was the same for respondents versus the general population, 27.5% in each case.

Annual household income distribution was notably similar to the general population for the lowest income group of under $30,000 (22.3% versus 22.1%). There were fewer respondents reporting incomes above $100,000 compared to the general population (24.7% vs. 33.6%), and more respondents in the two middle income brackets (roughly 26% vs. 22% for each).

The educational attainment of respondents tended to be higher than the general population with more completing some college or graduating from college (58.2% vs. 39.8% combining these two categories), but fewer completing graduate school (14.7% versus 18.1%). Fewer respondents were at the low end of the educational attainment distribution (some high school or high school graduate with 27.1% versus 42.2% combined categories for respondents compared to the general population).

The distribution of gender among respondents closely aligns with that of the general population, with roughly equal proportions of cis-females and cis-males. The transgender and gender independent group is also similar to the low end of the Census estimates (1.4% versus 1.6%).

While the distribution of race and ethnicity among respondents mostly reflects that of the general population, the proportion of Asian and Pacific Islander individuals is substantially lower compared to the general population (2.8% versus 6.5%). The proportions of several other race and ethnicity groups are slightly higher among respondents

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TABLE 1 Respondent characteristics compared to general U.S. population [unweighted sample].

Respondent attributes	Source	AmeriSpeak® panelists completing screening survey	U.S. adult (18+) population, 2020	
Patient age (adults)	1			
18–24 Years Old		9.8%	12.1%	
25–34 Years Old		17.7%	15.4%	
35–44 Years Old		15.5%	18.3%	
45–54 Years Old		13.9%	15.8%	
55–64 Years Old		18.4%	16.8%	
65–74 Years Old		16.7%	12.8%	
75 Years and Older		8.0%	8.8%	
Annual household income	5			
Under $30,000		22.3%	22.1%	
$30,000–$59,999		26.5%	22.0%	
$60,000–$99,999		26.6%	22.4%	
$100,000 and Above		24.7%	33.6%	
Education completed	4			
Some High School		7.3%	13.7%	
High School Grad		19.8%	28.5%	
Some College		38.8%	28.2%	
College Grad		19.4%	11.6%	
Graduate School		14.7%	18.1%	
Gender	2,3			
Cis-Female		49.6%	50.5%	
Cis-Male		49.0%	47.2%	
Transgender and Gender Independent		1.4%	1.6–2.3%	*
Race and ethnicity	4			
White non-Hispanic (NH)		62.5%	62%	
Black NH		13.1%	12%	
Asian and Pacific Isles, NH		2.8%	6.5%	
Other Race NH		1.2%	1%	
Multiple Races NH		2.7%	2%	
Hispanic (all races)		17.7%	17%	
Geographic location	5			
Urban/Metro		84.6%	80.0%	
Rural/Non-metro		15.4%	20.0%	
Health and employment status	5			
Working or Looking for Work		67.3%	63.4%	
Not in Labor Force, Nondisabled		27.0%	29.2%	
Disabled, Unable to Have Full-time Work		5.8%	7.4%	
Marital status	1			
Married		45.5%	53.0%	
Single, Divorced, Separated, Widowed		54.5%	47.0%	

1. American Community Survey (2021). 2. Pew Foundation. 3. Census Household Pulse Survey July 21–September 13, 2021. 4. National Health Interview Survey. 5. Shrider, EA, M Kollar, F Chen, J Semega, Current Population Reports, P60-273, Income and Poverty in the United States: 2020, U.S. Government Publishing Office, Washington, DC, September 2021. *Census questions for transgender and gender independent group are still in pilot testing. Percentages show range across wording.

compared to the general population (e.g., 17.7% versus 17% for Hispanics [all races], 13.1% versus 12% for Black, and 2.7% versus 2% for those reporting multiple races).

Although the majority of respondents reside in urban or metro areas, consistent with the distribution in the general population, there were fewer respondents from rural or non-metro locations (15.4% versus 20% in the general population). The distribution of health and employment status among respondents is largely comparable to that of the general population, with a somewhat lower proportion of individuals reporting disabled status (unable to have full-time work) among respondents compared to the general population (5.8% vs. 7.4%). The respondent sample had a smaller proportion of those currently married than did the general population, though in each case the sample was fairly evenly divided between those who were married and those not.

Overall, the comparison between respondent characteristics and the general U.S. adult population suggests that the sample captured a diverse and representative population, enhancing the generalizability of the study findings.

3.2 Illustrative examples of study outcomes

Table 2 presents narrative examples excerpted from patient and care partner reports to illustrate study outcome variables. Examples of diagnostic P&Ms include delays in diagnosis, problems with diagnostic testing, nonspecific diagnosis, and unresolved diagnosis. Examples of persisting physical harms reflecting severe impact of the diagnostic P&M include chronic pain, damage to extremities and nerves, and continuing functional limitations. Examples of persisting emotional harms indicative of severe impact of the diagnostic P&M include significant frustration, anger, feelings of invisibility, and stigmatization for patients reporting about themselves. Similarly, care partners experiences of persistent emotional harms include expressions such as "it broke my heart" and "it has affected the family and myself in ways words cannot express." Examples of a personal attribute or combination of personal attributes that impaired diagnosis and suggest diagnostic inequity include gender, too young an age for the diagnosis ultimately determined, being a person with a disability, weight ("because you are fat"), and being Latina.

3.3 Prevalence of diagnostic P&Ms and their effects

Among the 3,995 survey respondents in the household sample, Figure 1 shows that 37.7% reported experiencing at least one diagnostic P&M as a care partner or as a patient in the past 4 years, while 19.2% reported experiencing multiple P&Ms during the same period. Among respondents reporting on their own diagnostic P&Ms (only patients), 20.9% reported experiencing at least one diagnostic P&M in the past 4 years, while 10.3% reported experiencing multiple P&Ms during the same period. The population rate of perceiving personal attributes as impairing diagnosis was 6.6% of household respondents and 4.4% of patients themselves. Figure 1 also shows the prevalence of concerns about having a future diagnostic P&M being very likely in each sample.

3.4 Outcomes among respondents who experienced at least one P&M

Among the 1,506 patient and household care partner respondents reporting on diagnostic P&Ms within their household, Figure 2 shows that about 50% of respondents had experienced multiple P&Ms in the past 4 years. Based on responses for the selected P&M explored in detail in the survey, the longer-term effects resulting from that diagnostic P&M included persisting emotional harm (anxiety) and lasting physical harm. Among household respondents, about 29.2% reported persistent emotional harm, while 20.1% reported persistent physical harm of the person who experienced a P&M (Figure 2). Based on the subsample of patients reporting on themselves, 35.3% reported persisting anxiety, and 17.2% experienced persistent adverse physical effects. Almost 15% of those who experience a P&M thought it was very likely that they would experience another diagnostic P&M in the future.

Almost 22% of patients and 18.0% of the household respondents who reported a diagnostic P&M indicated that a personal attribute had played a role in their problematic diagnostic experience. The personal attributes associated with these pernicious effects sometimes corresponded to sociodemographic categories commonly used in social surveys and identified as sources of disparities in prior studies, but the diagnostic narratives also reported on more finely grained racial, ethnic and cultural identities or other characteristics (e.g., prior diagnosis of mental illness or substance use disorder, large body size). These self-reported attributes and their relationships to diagnostic experiences and outcomes will be examined separately in a forthcoming publication based on the open-ended narrative data.

3.5 Sociodemographic predictors of diagnostic P&Ms

Table 3 shows that regression results for both the household level (i.e., respondents reporting either about their own diagnosis or that of another person in their household) and patient subsample (i.e., patients reporting about their own diagnosis) analyses are similar in terms of significant sociodemographic predictors of P&Ms, though some results differ. We report results for each analytic frame separately since strategies for addressing diagnostic inequities may be targeted to individuals or households or both.

3.5.1 Household level analysis (full sample of all reported P&Ms)

At the household level, younger patients (18–24 years old) exhibited significantly higher odds of experiencing at least one diagnostic P&M (odds ratio 2.99, $p < 0.0001$) and multiple P&Ms (odds ratio 2.61, $p < 0.0001$) compared to those aged 45–54 years old. Conversely, older patients reported significantly lower odds of experiencing at least one P&M (odds ratio 0.62, $p = 0.001$ for those 65–74 years old and 0.64, $p = 0.012$ for 75 and older) and multiple P&Ms (odds ratio 0.56, $p = 0.004$ for the 65–74 group; and 0.46, $p = 0.006$ for those 75 and older) compared to the reference group (age 45 to 54).

Respondents from households with below average income (under $30,000 and $30,000–$59,999) exhibited significantly higher odds of experiencing at least one diagnostic P&M and multiple P&Ms compared to those from households with incomes of $100,000 and above. The odds ratios followed a consistently inverse gradient with

TABLE 2 Illustrative excerpts of outcome variables.

Outcome variable	Illustrative excerpt
Diagnostic P&M	Patient Report
	Issue with back. I thought disk was herniated but doctor refused to look into it. Was later diagnosed as a herniated disk almost 8 months later after more damage had been done. Zero treatments were effective in helping it…
	I went to the health clinic to get blood drawn to diagnose my potential thyroid problem. I do not know if it was the personnel at the clinic or the transporters that mishandled my samples, but they had to call me back to get more blood drawn because they could not get the information they needed to properly diagnose me from the first set of samples.
	Care Partner Report
	My father had a very bad rash (itching, blisters) that was first diagnosed by his PCP as just a rash. I searched on the internet and eventually came across Bullous Pemphigoid. After a second visit to the PCP, he agreed with me.
	My wife was having pain in her legs causing discomfort when walking… She did have a mass of blue veins that she thought might be the problem. She was told it was a dermatology problem. She later had some veins stripped from her legs. The problem continued. She had either two or three custom made lifts for her shoes, and then went to the "Good Feet Store" trying to get rid of the pain. In all cases, she was told that it should help. Nothing has helped, and she still has the problem which is getting worse.
Persisting Physical Harms → Diagnostic P&M with Severe Impact	Patient Report
	I had lots of breathing issues and chest pain at night… It was the third doctor who found the 5-inch-long tumor in my chest that was pressing on my lungs and causing pleurisy and pain… I wish I had requested imaging early on. All it took was a simple CT scan to diagnose the problem correctly, but it took almost a year and 3 doctors to get that scan. I was miserable for months and months. And after the surgery that removed the tumor, I was left with nerve damage and chronic pain that I still have that everyone tells me to just deal with. Overall, a horrible experience.
	The pain continued and worsened until belatedly doctors indicated surgery would be required. Because the situation had deteriorated so badly by then, the main nerves in my left leg became so damaged that they were no longer capable of normal use, and the operation failed to solve the problem. I now must use a walker at all times in order to stand or walk. My PCP has since admitted that an earlier surgery would have probably saved the leg.
	Care Partner Report
	[I wish I had known] that Guillan-Barre can take years to resolve and sometimes does not. What the other disabling factors are… They remained disabled with no definitive diagnosis.
	The doctors previously had been so damn preoccupied with the mysterious covid they could not be bothered with pneumonia everyone has known about and treated successfully for decades. He almost died because they ignored his lungs, his trouble breathing. All they cared about was covid… Almost 3 years later [he] is just now weaning himself off of extra oxygen to get through a normal day. He will never get back to the active life he lived before because his lungs are so damaged… Our lives will never be the same. We used to kayak when we camped. We cannot do that anymore. He does not have the strength. He cannot do a lot of the car maintenance he used to do. Very cold air, and very hot air bothers him. He just has to stop where before he could have kept doing what he was working on. It frustrates him sometimes.

(Continued)

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TABLE 2 (Continued)

Outcome variable	Illustrative excerpt
Persisting Emotional Harms → Diagnostic P&M with Severe Impact	Patient Report
	I had chronic productive coughing, and recurring lung infections. I was diagnosed with bronchitis 3 times, walking pneumonia once (for 4 different incidents of infection). Then it became too hard to work and deal with this. I barely slept. I could not stop coughing. I was then told I had RADS—Reactive airway disorder. I then sought out information about RADS and it was clear I did NOT have that. Then I was diagnosed with whooping cough, without giving me a pertussis test… just because I had uncontrollable coughing fits. I was FINALLY referred to a pulmonologist after an ER visit due to my inability to breathe. The pulmonologist thought it was uncontrolled allergies. I had an allergy test and started immunotherapy. That helped, but I still had infections and wheezing. I was finally diagnosed with asthma last July. The treatment for asthma has really helped me… I described my situation in detail. Providers pulled answers sometimes out of their asses. Tests were not performed to try to narrow down my problems, just to support their next hypothesis. I felt invisible, unheard, and very frustrated. I was devastated emotionally. My boss tried to performance manage me out of my job because I was worried more about staying alive than her pet projects. I had to reach out to several mental health therapists to cope with my frustration. I'm gray-haired and (at the time) approaching 60, and female. I am invisible. They just want me to go away.
	I've been bedridden 80% of the day for around 10 years. I'm considered a chronic pain patient, which is the "unsorted" bin where they toss those of us without a clear diagnosis… I'm on Medicaid (Title 19) and the program does not pay enough to make them care. When they could not figure out what was wrong, or why, my symptoms suddenly became "all in my head." They suggested a psychiatrist might be helpful. I suggested they should all go to hell and refused further visits. That's about where things stand presently. When I'm forced to interact with other doctors it gets ugly really quickly.
	Care Partner Report
	My daughter had her breast removed as she is high risk for breast cancer. Nobody, including her PCP who really was now responsible for her follow up care informed her about the fact that her bone density should be taken care of. She found out about 8 months after the surgery, her bones were bones of an 80-year-old and she is only 38 years old… She Cried a Lot. A Real Lot. It broke my heart. She is doing everything in her power to strengthen her bones and it has not been easy by any means. She works on it every day and it is strenuous.
	My grandmother had fallen and was bleeding from her ears. Family knew immediately she had a concussion. Took her to our town's local E.R. where doctors cleaned off the blood and said she did not have a concussion and sent her home. She was immediately acting strange and a couple days later [we] took her back to the E.R. because she was getting worse and that's when the family and my grandmother found out from the Radiologist and Neurologist that she had a level 4 concussion. She never recovered properly and passed away due to complications from the concussion… It has affected the family and myself in ways words cannot express. Mentally, emotionally, and physically.

(Continued)

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TABLE 2 (Continued)

Outcome variable	Illustrative excerpt
Personal Attribute(s) Perceived as Impairing Diagnosis → Diagnostic P&M Inequity	Patient Report
	Was misdiagnosed with an infection. I was then given incorrect medication which led to an allergic reaction… I was called names accused of being a difficult patient and because I am legally blind my competency was questioned. When I disagreed about the diagnosis and treatment the nurse questioned my competency. When I tried to involve administrators I was harassed. For a short time I refused to see any medical professional out of fear. During that time my condition worsened. I reached out to a mental health professional who encouraged me to do my own research and reach out to a new doctor… I lost the ability to walk. I had to drop out of my last semester of college. I isolated myself and was severely depressed. I refused to trust any medical professional. I ended up with irreversible joint damage. To this day I still have trust issues. Because of my experiences I only seek out care when things get really bad… I know there were factors such as my gender, being legally blind and my age that impacted my experience. Even though women are more likely to be diagnosed with an autoimmune disease we still live in the shadow of health care being designed around men. When you are younger than the typical person diagnosed you are not taken seriously. And being a person with a disability you are treated as though you cannot possibly be competent to make your own decisions. I am confident that if these factors were not present, things would have gone differently.
	I wish a doctor had taken all of my symptoms into thought instead of looking only at individual symptoms. All my symptoms taken together perfectly fit my actual diagnosis… I frequently received diagnoses that boiled down to "It's because you are fat." I'm very overweight and all my other symptoms were frequently ignored. In the end, weight gain was a symptom and not the problem.
	Care Partner Report
	His primary doc refused to biopsy a mass he had grown on his abdomen due to his age and the mass having characteristics of a lipoma. A year later they removed the mass due to its continued growth and it was found to be cancerous… His age impacted his diagnosis because since he was 30 they acted like it was impossible for him to have cancer.
	My wife injured herself at work and went to the hospital and they told her that she had a bruised muscle but ended up being her shoulder… They apologized and said that I was in my right to make a complain and that I did… I seen the difference in service from her being Latina and other races at the hospital.

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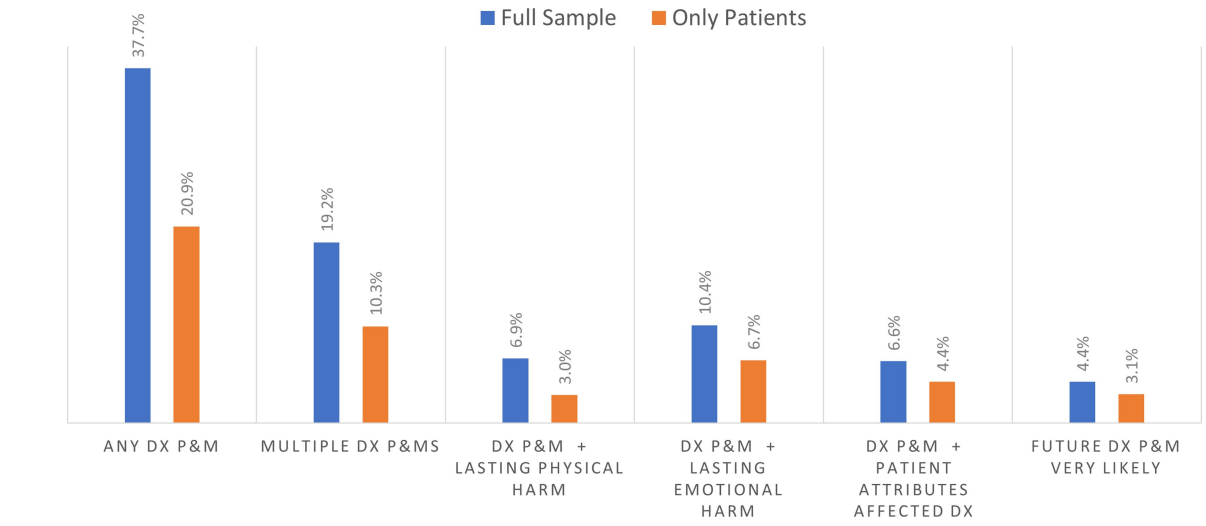

FIGURE 1
Prevalence of diagnostic problems and mistakes (DX and P&MS) in past 4 years and related outcomes.

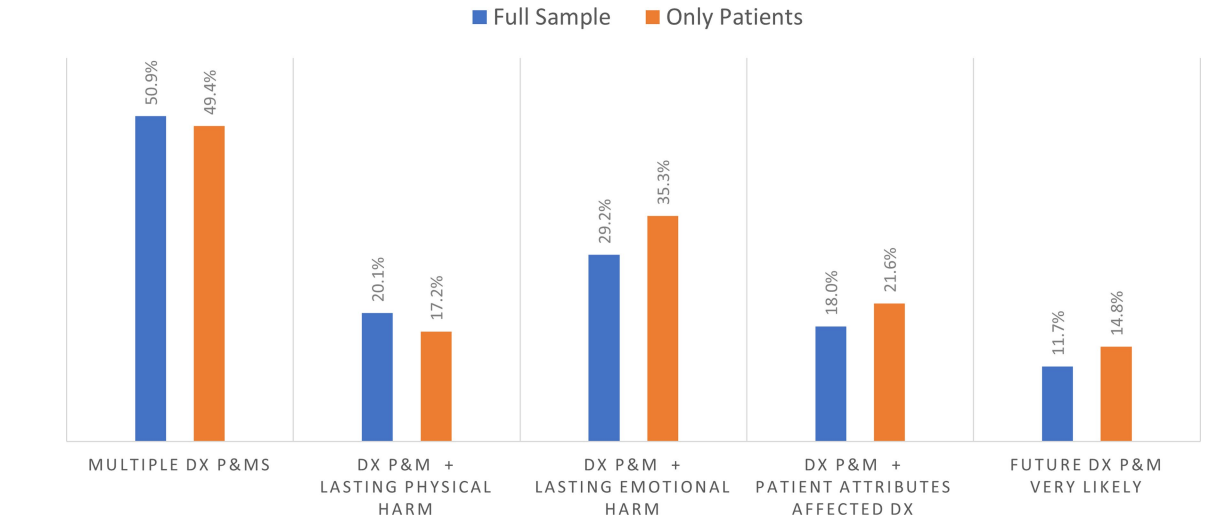

FIGURE 2
Percentage of outcome for those experiencing at least one diagnostic problem and mistake (DX and P&MS).

higher and more significant odds for P&Ms (at least one and multiple) as income levels decreased.

Respondents with less education completed, specifically those with only some high school education or high school graduation, exhibited significantly lower odds of experiencing at least one diagnostic P&M compared to those in the reference group of those who had completed some college (odds ratio 0.47, $p < 0.0001$ for some high school; odds ratio 0.72, $p < 0.003$ for high school graduation). Only the lowest educational attainment groups (some high school) reached statistical significance in predictions of multiple P&Ms, and similarly had lower odds compared to the reference group (odds ratio 0.54, $p = 0.005$).

Other significant sociodemographic predictors of increased risk of diagnostic P&Ms are related to gender, race and ethnicity, and disability. Cis-female respondents were significantly more likely to report experiencing any P&M (odds ratio 1.25, $p = 0.005$) and multiple P&Ms (odds ratio 1.28, $p = 0.02$) compared to cis-males. Transgender and gender independent individuals exhibited the highest risks: compared to cis-males (odds ratio 5.27, $p < 0.0001$ for at least one P&M and odds ratio 2.79, $p = 0.0004$ for multiple P&Ms). Disparity predictions for most racial and ethnic groups did not reach statistical significance. However, individuals identifying as multiracial exhibited significantly higher odds of P&Ms compared to non-Hispanic White individuals (odds ratio 1.80, $p = 0.007$ for at least one P&M; and odds ratio 1.77, p = 0.02 for multiple P&Ms). Individuals who identified their work status as disabled (unable to have full-time work) had significantly higher odds of experiencing diagnostic P&Ms compared to those in the reference group (odds ratio 1.94, $p < 0.0001$ for at least one P&M; and odds ratio 2.28, $p < 0.0001$ for multiple P&Ms).

Overall, the findings highlight the presence of disparities in the prevalence of diagnostic P&Ms within households, with significant associations observed for patient age, household income, education, gender, race and ethnicity, and disability status.

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TABLE 3 Disparities in prevalence of diagnostic P&Ms within respondent's household.

Respondent attributes	Frequency of diagnostic P&Ms		Frequency of diagnostic P&Ms with severe impact		Comparison group
	At Least One P&M (Problems + Mistakes)	Multiple P&Ms (in past 4 Years)	Persisting physical harm	Persisting emotional harm	NS (not significant) for $p > 0.05$
	Odds Ratio [prob]	Odds Ratio [prob]	Odds Ratio [prob]	Odds Ratio [prob]	
Patient age (adults)					Age 45–54
18–24 Years Old	2.99 [<0.0001]	2.61 [<0.0001]	1.08 [NS]	1.83 [0.01]	
25–34 Years Old	1.23 [NS]	1.17 [NS]	0.83 [NS]	1.27 [NS]	
35–44 Years Old	1.07 [NS]	1.10 [NS]	1.23 [NS]	1.12 [NS]	
55–64 Years Old	0.88 [NS]	0.71 [NS]	0.71 [NS]	0.72 [NS]	
65–74 Years Old	0.62 [0.001]	0.56 [0.004]	0.55 [NS]	0.48 [0.004]	
75 and Older	0.64 [0.02]	0.46 [0.006]	0.60 [NS]	0.41 [0.01]	
Household income					$100,000 and More
Under $30,000	1.45 [0.005]	1.93 [0.0001]	1.58 [NS]	1.94 [0.002]	
$30,000–$59.999	1.32 [0.02]	1.69 [0.001]	1.75 [0.02]	1.71 [0.006]	
$60.000–$99,999	1.13 [NS]	1.29 [NS]	0.93 [NS]	1.43 [NS]	
Education completed					Some College (inc. AA degree)
Some High School	0.47 [<0.0001]	0.54 [0.005]	0.54 [NS]	0.71 [NS]	
High School Grad	0.72 [0.003]	1.00 [NS]	0.86 [NS]	0.81 [NS]	
College Grad	0.94 [NS]	1.03 [NS]	1.26 [NS]	0.93 [NS]	
Graduate School	1.03 [NS]	1.07 [NS]	0.89 [NS]	0.99 [NS]	
Gender					Cis-Male
Cis-Female	1.25 [0.005]	1.28 [0.02]	1.45 [0.03]	1.36 [0.01]	
Transgender and Gender Independent	5.27 [<0.0001]	2.79 [0.0004]	2.29 [NS]	1.94 [NS]	
Race and ethnicity					White, non-Hispanic
Black, NH	1.03 [NS]	1.00 [NS]	0.64 [NS]	0.61 [NS]	
Hispanic (all races)	1.07 [NS]	1.14 [NS]	0.95 [NS]	0.90 [NS]	
Asian and Pacific Isles, NH	0.73 [NS]	1.23 [NS]	0.46 [NS]	0.61 [NS]	
Other Race, NH	1.03 [NS]	0.29 [NS]	--	0.68 [NS]	
Multiple Races, NH	1.80 [0.007]	1.77 [0.02]	2.17 [0.03]	1.68 [NS]	
Location					Rural/Outside metropolitan
Urban/Metro	0.84 [NS]	0.93 [NS]	0.89 [NS]	1.00 [NS]	
Work status					Not Work-Disabled
Disabled, Unable to Have FT Work	1.94 [<0.0001]	2.28 [<0.0001]	2.76 [0.0001]	2.22 [0.0002]	
Marriage status					Not married
Married	1.03 [NS]	0.90 [NS]	0.89 [NS]	0.97 [NS]	

NH, non-Hispanic; FT, Full-time; NS, not significant.

3.5.2 Patient-level analysis (subsample reporting about their own diagnosis)

Table 3 also shows the regression analysis for patients reporting about themselves. As with the household analysis, there were no significant differences in the prevalence of diagnostic P&Ms between respondents residing in urban/metro areas compared to those in rural/non-metro areas. Nor were disparities predicted based on marital status (married versus not married).

Both patient age and household income predictions remained similar to the household analysis as expected, given that these sociodemographic characteristics are consistently identified in both samples (Exhibit 2). Patient age was a significant predictor of diagnostic

P&Ms, both at least one and multiple P&Ms in the past 4 years. Younger patients (under 25) exhibited significantly higher odds of experiencing at least one diagnostic P&M (odds ratio 3.98, $p < 0.0001$) and multiple P&Ms (odds ratio 3.48, $p < 0.0001$) compared to those aged 45–54. Both these point estimates were higher than in the household analysis. Conversely, older patients (65 and older) had significantly lower odds of experiencing both at least one P&M (odds ratio 0.60, $p = 0.006$ for those 65 to 74; odds ratio 0.58, $p = 0.03$ for those 75 and older) and multiple P&Ms (odds ratio 0.48, $p = 0.005$ for those 65 to 74; odds ratio 0.43, $p = 0.02$ for 75 and older), compared to the reference group.

Respondents with below average incomes again reported more diagnostic P&Ms. Those living in households with the lowest incomes (under $30,000) exhibited significantly higher odds of experiencing P&Ms compared to those from households with incomes of $100,000 and above (odds ratio 1.59, $p = 0.003$ for at least one P&M; odds ratio 2.39, $p < 0.0001$ for multiple P&Ms). For those with household incomes in the next lowest bracket of $30,000–$59,999, respondents reporting about themselves also had significantly greater risk of at least one P&M (odds ratio 1.35, $p = 0.03$) and multiple P&Ms (odds ratio 1.81, $p = 0.002$).

Respondents with lower levels of education, specifically those with only some high school education or high school graduation, exhibited significantly lower odds of experiencing at least one diagnostic P&M compared to the reference group of those who had some college (odds ratio 0.30, $p < 0.0001$ for some high school; odds ratio 0.56, $p < 0.0001$ for high school graduation). Those with some high school also had significantly lower odds of multiple P&Ms (0.37, $p = 0.003$). The differences in point estimates were more pronounced at the patient level compared to the household analysis.

Cis-female respondents had significantly higher odds of experiencing diagnostic P&Ms compared to cis-male respondents (odds ratio 1.2, $p = 0.04$ for at least one P&M; odds ratio 1.35, $p = 0.02$ for multiple P&Ms). Transgender and gender independent individuals also exhibited significantly higher odds of experiencing at least one P&M compared to cis-males (odds ratio 2.04, $p = 0.05$). Although the direction of the effects in the patient analysis were again consistent with the household analysis, the point estimates indicated either the same or less separation from the reference group.

As with the household analysis of racial and ethnic predictors, only individuals identifying as multiracial exhibited significantly higher odds of experiencing P&Ms compared to non-Hispanic White individuals (odds ratio 1.80, $p = 0.03$ for at least one; odds ratio 2.00, $p = 0.03$ for multiple P&Ms). Point estimates in the household and patient analyses were quite similar.

Disabled (unable to have full-time work) respondents had significantly higher odds of experiencing diagnostic P&Ms compared to the reference group (odds ratio 2.27, $p < 0.0001$ for at least one; odds ratio 2.66, $p < 0.0001$ for multiple P&Ms).

Overall, the findings highlight the presence of disparities in the prevalence of diagnostic P&Ms affecting respondents' own healthcare, with significant associations observed for patient age, household income, education completed, gender, race and ethnicity, and disability in both analyses (household and patient only).

3.6 Persistent harms from diagnostic P&Ms

Table 4 presents the regression results of sociodemographic predictors of diagnostic P&Ms with severe impacts for both the full household sample and the patient subsample. While the direction of effects of sociodemographic predictors by subgroups is similar in both analyses, there are some predictors that are only statistically significant in one of the analyses.

3.6.1 Household level analysis

As shown in the Table 4 household regression results, being cis-female or disabled (unable to have full-time work) was associated with significantly higher odds of both persistent emotional harm (anxiety of the respondent) and persistent physical harm (for the patient) compared to the reference group. For emotional harm, being cis-female increased odds to 1.36 ($p = 0.01$) and being disabled (unable to have full-time work) increased odds to 2.22 ($p = 0.0002$) compared to the reference groups. Being cis-female, disabled or multi-racial was also associated with significantly higher frequencies of persistent physical harm, with odds ratios of 1.45 ($p = 0.03$); 2.76 ($p = 0.0001$); and 2.17 ($p = 0.03$), respectively.

For the lowest income group (under $30,000), only persisting emotional harms exhibited statistically significant increased likelihood (odds ratio 1.94, $p = 0.002$) compared to the highest household bracket of $100,000 or more. Increased odds of persisting physical and emotional harm were each significant for the $30,000 to 59,999 group (odds ratio 1.75, $p = 0.02$; and odds ratio 1.71, $p = 0.006$, respectively) compared to the reference group. Respondents reporting on patients in the lowest age group (18 through 24 years old) had significantly higher odds of persisting emotional harm (odds ratio 1.83, $p = 0.01$).

Significant predictors of decreased odds of persisting emotional harm after diagnostic P&Ms were seen for older age groups (odds ratio 0.48, $p = 0.004$ for those 65 to 74 years old and 0.41, $p = 0.01$ for those 75 and older).

3.6.2 Patient level analysis

Similar to the household level regression analysis results, the odds of higher frequencies of persisting harm (physical or emotional) were significant for lower income, cis-female, and disabled (unable to have full-time work) work status (Table 4). The highest and most significant odds of both persisting physical and emotional harms were experienced in the disabled (unable to have full-time work) group compared to the reference group (odds ratio 4.11, $p < 0.0001$ for physical harm; odds ratio 2.55, $p = 0.0002$ for emotional harm). Significantly lower odds for older adults for persisting emotional harms were also present in the patient subsample.

Unlike the household sample, Table 4 shows higher prevalence of persisting physical harms for the 65–74 years old age group compared to those 45 to 54 (odds ratio 0.26, $p = 0.006$). Lower educational attainment significantly predicted reduced odds of physical harm for those with some high school (odds ratio 0.37, $p = 0.05$), and reduced emotional harm for high school graduates (odds ratio 0.66, $p = 0.04$) compared to the reference group.

The significant race and ethnicity predictors were not the same in the patient subsample. Patients who identified as Black, non-Hispanic were also at significantly lower risk of reporting persisting physical harm about oneself (odds ratio 0.41, $p = 0.03$ compared to reference group), as well as persisting emotional harm (odds ratio 0.46, $p = 0.004$). The disparity in prevalence of statistically significant higher odds of persisting physical harm noted for those identifying as multiracial in the household sample was not seen in the patient subsample (odds ratio 1.19 [not significant] versus 2.17, $p = 0.03$ in the household analysis).

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TABLE 4 Disparities in prevalence of diagnostic P&Ms affecting respondents' own health care.

Respondent attributes	Frequency of diagnostic P&Ms		Frequency of diagnostic P&Ms with severe impact		Comparison group
	At Least One P&M (Problems + Mistakes)	Multiple P&Ms (In Past 4)	Persisting physical harm	Persisting emotional harm	NS (not significant) for $p > 0.05$
	Odds Ratio [prob]	Odds Ratio [prob]	Odds ratio [prob]	Odds ratio [prob]	
Patient Age (Adults)					Age 45–54
18–24 Years Old	3.98 [<0.0001]	3.48 [<0.0001]	1.91 [NS]	2.26 [0.003]	
25–34 Years Old	1.50 [NS]	1.37 [NS]	0.95 [NS]	1.33 [NS]	
35–44 Years Old	1.24 [NS]	1.23 [NS]	1.30 [NS]	0.95 [NS]	
55–64 Years Old	0.99 [NS]	0.82 [NS]	0.60 [NS]	0.76 [NS]	
65–74 Years Old	0.60 [0.006]	0.48 [0.005]	0.26 [0.006]	0.34 [0.001]	
75 and Older	0.58 [0.03]	0.43 [0.02]	0.31 [NS]	0.31 [0.01]	
Household Income					$100,000 and More
Under $30,000	1.59 [0.003]	2.39 [<0.0001]	1.83 [NS]	2.00 [0.005]	
$30,000–$59.999	1.35 [0.03]	1.81 [0.002]	2.10 [0.03]	1.77 [0.01]	
$60.000–$99,999	1.09 [NS]	1.39 [NS]	1.04 [NS]	1.55 [NS]	
Education Completed					Some College (inc. AA degree)
Some High School	0.30 [<0.0001]	0.37 [0.003]	0.37 [0.05]	0.65 [NS]	
High School Grad	0.56 [<0.0001]	0.75 [NS]	0.61 [NS]	0.66 [0.04]	
College Grad	0.89 [NS]	0.92 [NS]	0.86 [NS]	0.83 [NS]	
Graduate School	1.02 [NS]	1.00 [NS]	0.71 [NS]	1.00 [NS]	
Gender					Cis-Male
Cis-Female	1.22 [0.04]	1.35 [0.02]	1.81 [0.008]	1.55 [0.003]	
Transgender and Gender Independent	2.04 [0.05]	1.87 [NS]	0.89 [NS]	1.68 [NS]	
Race and Ethnicity					White, Non-Hispanic
Black, NH	1.02 [NS]	1.00 [NS]	0.41 [0.03]	0.46 [0.004]	
Hispanic (all races)	0.95 [NS]	1.02 [NS]	0.77 [NS]	0.77 [NS]	
Asian and Pacific Isles, NH	0.95 [NS]	1.66 [NS]	0.74 [NS]	0.58 [NS]	
Other Race, NH	1.15 [NS]	--	--	0.52 [NS]	
Multiple Races, NH	1.80 [0.03]	2.00 [0.03]	1.19 [NS]	1.70 [NS]	
Location					Rural/Outside metropolitan
Urban/Metro	0.92 [NS]	0.92 [NS]	0.79 [NS]	1.06 [NS]	
Work Status					Not Work-Disabled
Disabled, Unable to Have FT Work	2.27 [$p < 0.0001$]	2.66 [<0.0001]	4.11 [<0.0001]	2.55 [0.0002]	
Marriage Status					Not married
Married	1.06 [NS]	0.99 [NS]	0.95 [NS]	0.85 [NS]	

NH, non-Hispanic; FT, Full-time; NS, not significant.

3.7 Diagnosis impaired by personal attributes: household and patient-level analyses

Table 5 displays regression results for both analytic frames. For respondents in both samples, the youngest patient group (18–24 years old) was twice as likely to endorse perceiving that a personal attribute impaired diagnosis compared to the reference group (odds ratio 1.84, $p = 0.04$ for household sample; odds ratio 2.02, p = 0.04 for patient sample). The two oldest age categories had significantly lower odds of perceiving that a personal attribute impaired diagnosis with the lowest odds for patients 75 and older.

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TABLE 5 Personal attribute perceived as impairing diagnosis during diagnostic P&M.

Respondent attributes	Likelihood of personal attribute effect				Comparison group
	Household analysis		Patients reporting on themselves		NS (not significant) for $p > 0.05$
	Odds Ratio	prob	Odds Ratio	prob	
Patient Age (Adults)					Age 45–54
18–24 Years Old	1.84	0.04	2.02	0.04	
25–34 Years Old	0.99	NS	1.19	NS	
35–44 Years Old	0.83	NS	0.86	NS	
55–64 Years Old	0.69	NS	0.73	NS	
65–74 Years Old	0.42	0.005	0.38	0.008	
75 and Older	0.44	0.05	0.28	0.02	
Household Income					$100,000 and More
Under $30,000	1.83	0.02	1.97	0.03	
$30,000–$59.999	1.79	0.02	1.69	NS	
$60.000–$99,999	1.29	NS	1.33	NS	
Education Completed					Some College (inc. AA degree)
Some High School	0.22	0.002	0.10	0.002	
High School Grad	0.65	0.05	0.41	0.001	
College Grad	1.28	NS	1.05	NS	
Graduate School	1.15	NS	1.20	NS	
Gender					Cis-Male
Cis-Female	2.12	<0.0001	2.48	<0.0001	
Transgender and Gender Independent	5.36	<0.0001	5.64	<0.0001	
Race and Ethnicity					White, Non-Hispanic
Black, NH	0.83	NS	0.78	NS	
Hispanic (all races)	0.90	NS	0.83	NS	
Asian and Pacific Isles, NH	0.62	NS	0.22	NS	
Other Race, NH	0.80	NS	1.07	NS	
Multiple Races, NH	1.77	NS	1.40	NS	
Location					Rural/Outside metropolitan
Urban/Metro	0.85	NS	1.02	NS	
Work Status					Not Work-Disabled
Disabled, Unable to Have FT Work	2.48	0.001	2.31	0.009	
Marriage Status					Not Married
Married	0.84	NS	0.87	NS	

NH, non-Hispanic; FT, Full-time; NS, not significant.

In both the household and patient samples, higher odds of experiencing an impaired diagnosis based on personal attributes were significantly predicted at almost twice the odds for the lowest income brackets (under $30,000) as well as for the next lowest bracket ($30,000–$59,999) for the household sample compared to reference group ($100,000 and more). Lower levels of educational attainment significantly predicted much lower likelihood of perceived personal attribute effect, with the lowest odds reported by patients themselves with some high school (odds ratio 0.10, $p = 0.002$) compared to the reference group (some college education).

Higher rates of perceiving a personal attribute impaired diagnosis were predicted for cis-female gender and transgender and gender independent groups compared to the cis-male group in both samples (odds ratio 2.48, $p < 0.0001$ and odds ratio of 5.64, $p < 0.0001$, respectively). Inability to work due to a disability was also consistently associated with an elevated rate of reporting that diagnosis had been disrupted by a personal attribute.

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TABLE 6 Disparities in expectations for future diagnostic risks.

Respondent attributes	Likelihood of future diagnostic P&M				Comparison group
	Household analysis		Patients reporting on themselves		NS (not significant) for $p > 0.05$
	Odds Ratio	prob	Odds Ratio	prob	
Patient Age (Adults)					Age 45–54
18–24 Years Old	1.11	NS	0.90	NS	
25–34 Years Old	0.87	NS	0.73	NS	
35–44 Years Old	1.15	NS	0.93	NS	
55–64 Years Old	0.84	NS	0.79	NS	
65–74 Years Old	0.57	0.02	0.49	0.02	
75 and Older	0.57	NS	0.42	0.03	
Household Income					$100,000 and More
Under $30,000	1.18	NS	1.19	NS	
$30,000–$59.999	1.26	NS	1.34	NS	
$60.000–$99,999	1.08	NS	1.26	NS	
Education Completed					Some College (inc. AA degree)
Some High School	0.99	NS	1.16	NS	
High School Grad	1.04	NS	1.06	NS	
College Grad	1.08	NS	0.93	NS	
Graduate School	1.21	NS	1.06	NS	
Gender					Cis-Male
Cis-Female	1.30	0.03	1.60	0.002	
Transgender and Gender Independent	1.33	NS	3.34	0.01	
Race and Ethnicity					White, Non-Hispanic
Black, NH	0.76	NS	0.77	NS	
Hispanic (all races)	0.73	NS	0.87	NS	
Asian and Pacific Isles, NH	0.98	NS	1.27	NS	
Other Race, NH	0.88	NS	0.76	NS	
Multiple Races, NH	1.57	NS	1.25	NS	
Location					Rural/Outside metropolitan
Urban/Metro	1.37	0.05	1.10	NS	
Work Status					Not Work-Disabled
Disabled, Unable to Have FT Work	1.24	NS	1.19	NS	
Marriage Status					Not Married
Married	0.81	NS	0.82	NS	

NH, non-Hispanic; FT, Full-time; NS, not significant.

3.8 Expectations for future diagnostic risks

Table 6 illustrates several disparities in expectations for future diagnostic risks among respondents, as assessed by the likelihood of the respondent anticipating a future diagnostic P&M occurring when receiving health care. We report both household and patient-level analyses in the table side-by-side.

Older age groups had significantly lower odds of expecting a future diagnostic P&M compared to the reference group (odds ratio 0.49, $p = 0.02$ for 65–74; odds ratio 0.42, $p = 0.03$ for 75 and older for patients reporting on themselves). A similar pattern holds for the household level data, which include expectations reported by care partners. Within this sample, only the 65 to 74 group had statistically significantly lower odds of concern (odds ratio 0.57, p = 0.02).

In comparison to cis-males, cis-females and transgender and gender independent individuals who had experienced a diagnostic P&M had significantly higher odds of expecting future diagnostic P&Ms (odds ratio 1.60, $p = 0.0002$ for cis-females; odds ratio 3.34, $p = 0.01$ for transgender and gender independent in the household analysis). In the household sample, the cis-female group, but not the

transgender and gender independent group, had statistically significant higher odds of concern.

In both cases, the gender-related differences in future risk were consistent with the differences in P&M and harm experiences reported in Tables 3, 4. By contrast, households in urban/metro areas reported statistically higher odds of concern about future diagnostic P&Ms (odds ratio 1.37, $p = 0.05$). though these elevated risk perceptions were not matched by any comparable geographic differences in the experience of diagnostic P&Ms or harms.

4 Discussion

Our study aimed to fill several noteworthy gaps in the literature on diagnostic safety. First, it enriches our understanding of patient-reported diagnostic P&Ms by augmenting earlier findings estimating the national prevalence of harmful diagnostic events by incorporating multiple P&Ms, harmful consequences, and P&Ms attributable to differential treatment based on identified personal attributes. These new findings offer valuable insights into the prevalence of diagnostic breakdowns and their distribution across various sociodemographic groups, shedding light on disparities that may exist in healthcare experiences and outcomes related to diagnosis. The consistency of findings across multiple outcomes increases our confidence that these at-risk groups merit greater attention and protections.

Second, we successfully demonstrated the feasibility of obtaining patient-reported data from a national sample to better understand diagnostic P&Ms and their sociodemographic predictors. This includes responses from population subgroups that have historically had limited opportunities to voice problems and mistakes during their diagnostic experiences. And it includes data from narrative accounts that illuminate interactions within the diagnostic process in ways not previously visible to researchers.

Our analyses revealed several types of noteworthy findings that we group into three clusters. The first set involves results that are broadly consistent with findings from past studies, but which highlight nuances not identified in previous research. The second set of findings illuminate new sources of disparities for which we have not previously had reliable national estimates of magnitude, and fresh aspects or perspectives that more fundamentally alter how we should think about or address diagnostic inequities. The final cluster is in some ways the most generative, raising a variety of questions or puzzles that merit attention in future research.

In discussing our findings, we utilize definitions of health equity, diagnostic equity, health disparities and diagnostic disparities summarized in the public briefing book for the National Academies Workshop: "Advancing Equity in Diagnostic Excellence to Reduce Health Disparities." (46) Specifically, health equity is "the state in which everyone has a fair and just opportunity to attain their highest level of health." (47), while diagnostic equity is defined as "providing everyone with a fair and just chance of receiving a timely, accurate diagnosis to lead to appropriate interventions and health benefits, regardless of personal characteristics." (5, 20, 48) Similarly, health disparities are defined as "preventable differences in the burden of disease, injury, violence, or opportunities," (49) and "diagnostic disparities occur when diagnostic errors are experienced at disproportionate rates by certain patient subgroups based, for example, on patients' age, sex/gender, or race/ethnicity." (50) In our study, diagnostic disparities reflect experiences of problems and/or mistakes during a patient's diagnostic journey (diagnostic P&Ms), which may or may not be classified as diagnostic errors from a clinical point of view.

4.1 Better understanding previously documented diagnostic disparities

Our findings are largely consistent with those in the literature identifying elevated risk of diagnostic difficulties for young adults, cis-women, those living in low-income households and people with disabilities. In each case, however, the findings reported above highlight some implications that have been overlooked or downplayed in past research.

Younger adult patients, particularly those under 25 years old, experienced significantly higher rates of diagnostic P&Ms compared to their older counterparts. While prior studies have pointed to risks of delayed or missed diagnosis for younger people for specific clinical conditions (e.g., stroke, young adult cancers) (51, 52) and patient or clinician perceptions of the patient "being too young" for the diagnosis they ultimately received (41, 53–55), the population-based estimates of double to triple the chance of diagnostic P&Ms for this younger age group in our multivariate analysis suggests a need for bringing greater attention to both clinical and non-clinical contributors of this elevated risk.

Previous research has documented gender-related biases in diagnosis related to cis-women compared to cis-men, most commonly in terms of clinicians' dismissal of symptoms reported by patients (56, 57). These prior findings are echoed most strongly in our findings reported in Table 5, which highlights gendered differences that respondents observed in their interactions during diagnosis. But the elevated rates of diagnostic risk for cis-female respondents are also evident for persistent harms in the aftermath of diagnostic breakdowns. This could reflect a second-stage of dismissal, if cis-women's reports of symptoms related to diagnostic P&M itself are also taken less seriously than are comparable reports from cis-men.

Lower household income also emerged as a significant predictor of higher prevalence of diagnostic P&Ms, with the lowest income group facing the highest risks. This association is consistent with multiple qualitative and quantitative studies that single-out economic disadvantage as a predictor of diagnostic breakdowns (12, 14). But previous research involves samples too small to distinguish the scope of these financial risks. Our findings suggest that the scope is quite extensive—with all Americans living in households with below-average income experiencing elevated risks of diagnostic P&Ms. Developing effective interventions to mitigate the impact of financial barriers on diagnostic accuracy and timeliness likely will depend on close attention to dynamics both inside and outside of the medical system, as well to difficulties at the boundary of these two terrains that people must navigate as they become patients during a diagnostic process (5).

Previous research has also identified a variety of ways in which physical disabilities impair testing and other aspects of the diagnostic process (58). Because our analyses relied on the identification of disability through work status (disabled and unable to have full-time work), it suggests an alternative or additional pathway for increased

diagnostic risks in part due to different levels of connection to medical care or less extensive support with clinical issues from workplace human resource departments. Because disability can affect employment, social status, and sources of insurance in this way, our findings underscore the importance of recognizing and addressing the unique needs of disabled individuals within healthcare systems that go beyond clinical interactions, emphasizing the imperative for tailored interventions and supports. Interventions could also be developed based on analogous efforts in other targeted areas such as food insecurity for those who have disability (whether related to work status or not) to apply best practices for accessibility, universal design, and maximize input from the disability rights community (59, 60).

4.2 Newly identified aspects of diagnostic disparities

Our analysis identified other sociodemographic predictors of diagnostic P&Ms, affecting individuals who self-identify as transgender and gender independent, as well as those who identify as multiracial. Both groups were associated with substantially increased risk of experiencing diagnostic P&Ms and associated harms. But neither has received much attention in past research, despite their strikingly elevated risks.

In both cases, this situation reflects a common reluctance among researchers to report statistical results for subgroups that represent a relatively small portion of the American public. In fact, many studies explicitly suppress findings for subsamples that fall below an arbitrary size threshold (56). Consequently, smaller groups like those identifying as transgender or gender independent or those identifying as multiracial (both representing 2–3% of the American public) do not get reported in results, no matter how large the cross-group differences are in diagnostic or other health-related experiences.

This practice rests on inadequate statistical reasoning. To be sure, if sample sizes are small, the standard errors on the regression coefficients get inflated, and even large cross group differences may sometimes be statistically insignificant. (Note, for example, the nonsignificant but large odds-ratios on persisting harms for the transgender and gender independent respondents in Table 3. Or observe the persisting emotional harms for multiracial respondents in that same table.) But when comparisons remain statistically significant despite the small sample sizes, they often illuminate strikingly pronounced disparities, as can be observed for the transgender and gender independent respondents in Table 5. These should not be ignored.

A second set of new findings reflect subgroups of respondents who report substantially *fewer* diagnostic P&Ms or harms than the average patient. Here again, this is evident for two sets of respondents: those with more limited education and those over the age of 65. Consider first individuals with lower levels of education, particularly those with some high school education or having completed high school. Past statistical studies of diagnostic P&Ms have typically included either measures of education *or* measures of household income, but not both. Because our findings reveal that low-income is associated with increased risks, but lower education is associated with lower reported diagnostic P&Ms, failing to include both variables means that the two relationships would statistically cancel each other out, making it appear that lower socioeconomic status has no strong relationship to diagnostic outcomes at all.

Our finding, by contrast, thus opens space to hypothesize about why these offsetting associations exist. Perhaps individuals with lower education levels face fewer diagnostic problems compared to those with higher levels of education, though that seems unlikely. Alternatively, it may be that they are significantly less likely to report these effects. The challenges of adjusting to the complex terminology and terrain of health care among individuals with limited education, especially in the diagnostic stage of care, may make it harder to recognize and report diagnostic P&Ms (14, 28), resulting in underestimation of their prevalence as well as their impact. Alternatively, individuals with lower education levels may have developed lower expectations for healthcare, potentially leading them to be less likely to report deficits in care or attribute harms to diagnostic P&Ms. Further research is needed to explore the complex interplay between education level, healthcare expectations, health literacy, and diagnostic outcomes to inform strategies for improving healthcare quality and equity across diverse socioeconomic backgrounds.

As reported in our findings above, older adults, aged 65 and above, consistently demonstrated reduced odds of diagnostic P&Ms and harms. This might seem surprising, given the multiple comorbidities and polypharmacy that increase as people age, increasing the exposure to health care and any iatrogenic risks. However, one study in the UK found that older adults were more likely to have both higher expectations and be more satisfied with their care compared to younger patients (61). As expectations are socially constructed to a large degree, one's generational context (e.g., life as a "baby boomer"), as well as one's prior experiences within a given country's health system, could be relevant and potentially produce different patterns of expectations and reporting by patients and their care partners about health care experiences. Alternatively, the more stable and health-promoting coverage of the Medicare program may facilitate more regular visits to clinicians and thus more timely diagnoses among older Americans.

Finally, our study is the first to identify disparities in expectations regarding future diagnostic risks. Certain subgroups, including cis-females, and transgender and gender independent individuals particularly express heightened concerns. These findings emphasize the need for proactive measures to address patient anxieties about future care, improve communication, and address trust breaches between patients and healthcare providers, including interventions aimed at acknowledgement and repair. Additionally, more directed attention to how patients and their care partners reflect on their diagnostic P&M experiences and outcomes could deepen considerations about different ways that concerns about their future care could manifest (30).

4.3 Further puzzles and priorities for future research

Our findings illuminate a number of patterns among experiences and expectations regarding diagnostic disparities that merit additional attention from scholars and additional prioritization among funders of medical and health services research. We describe here five puzzling results that seem particularly deserving of future scrutiny.

First, there are some noteworthy differences in the relationship between experiences with diagnostic P&Ms (Tables 3–5) and expectations regarding future risks (Table 6). Gendered differences in risk of P&Ms and harms are matched by elevated concerns about future risk among cis-women and transgender or gender independent respondents. But other subgroups experiencing equally elevated

P&Ms—such as respondents with disabilities who are unable to have full-time work or those living in households with below-average income—do not appear to translate those experiences into elevated perceptions of future risk. Similar inconsistencies emerge for those reporting fewer diagnostic risks. Older Americans' perceptions of below-average diagnostic P&Ms and harms are matched by their expectations of lower future risks. But a comparable consistency of reduced experiences and expectations does *not* carry over to respondents with limited education. Better understanding the origins of these inconsistencies might offer useful insights into how people understand or interpret their past diagnostic experiences, their future expectations, or both.

Second, as noted above, the association between household income and diagnostic risks extends over a surprisingly large portion of the public. Authors of past studies have inferred that there might be a relationship between Medicaid coverage, reduced reimbursement rates for clinician visits, limited time spent in diagnosis, and consequently, elevated risk of diagnostic breakdowns (7, 12). But Medicaid coverage for adults is limited almost exclusively to those in the bottom quartile of the income distribution. Since elevated diagnostic risks and harms extends to the bottom *two* quartiles, some other causal or associative pathway must be in play. Research is needed to identify what that entails.

Third, our findings suggest that there is close congruence between subgroups that report identified diagnostic risk (individual or multiple P&Ms), diagnostic harms (persisting physical or emotional distress) and perceptions that patients were diagnosed differently and sub-optimally based on some identifiable personal attribute. What sort of interactions lead patients or care partners to make these attributions? And how are they able to discern this differential diagnostic process, when they are only observing their own or a care partner's diagnosis and not the experiences of other patients they do not know? These questions merit additional study.

Fourth, how might the perception that patients have been treated differently during diagnosis because of some personal attribute alter patients' (or care partners') longer-term relationships with individual clinicians or with the healthcare system as a whole? The excerpts in Table 2 highlight these perceptions, such as the respondent who stated: "And being a person with a disability you are treated as though you cannot possibly be competent to make your own decisions. I am confident that if these factors were not present, things would have gone differently." Is perceived discrimination, in particular, corrosive to trust in medical care or in health care professionals or both? Are there ways in which more positive expectations might be restored, despite a perception of past discrimination or other issues raised by these respondents? Here again, additional research is needed to address these questions.

Finally, contrary to expectation, a set of null findings is particularly vexing. In our multivariate analyses, except for predictors related to the multiracial group, other race and ethnicity groups did not emerge as a significant predictor of elevated diagnostic P&Ms or associated outcomes. These findings may reflect the complex and intersectional nature of healthcare disparities, where the influence of race and ethnicity on diagnostic outcomes may be mediated by other factors such as socioeconomic status. However, prior literature suggests grave inequities among racial and ethnic minorities arising from structural barriers, implicit bias, overt racism, and differential access to high-quality care (21, 62). It is vital to highlight that the statistical meaning of a null finding is not proof of no effect. Future research with larger samples will allow interaction analysis with race and ethnicity categories to further explore associations with diagnostic P&Ms and harms. At the same time, it is also possible that other explanations (e.g., concerns and resulting hesitations about reporting problems related to health care) deserve more attention in future studies of diagnostic P&Ms. For example, a scoping review found evidence of underreporting by clinicians of patient safety events for Black patients compared to White patients in voluntary reporting systems, which could correspond to biases in information supplied directly to Black patients and their care partners about what went wrong in their care (63).

4.4 The broader context of inequities based on other U.S.-based surveys

That diagnostic shortfalls perceived by patients and their families are unevenly distributed in the U.S. should, in itself, be unsurprising. Past surveys have long documented persisting inequities in Americans' reported economic insecurity (64), social anxieties (65, 66), and stigma related to various health conditions (67). Surveys of Americans' experiences within health care have similarly documented multiple inequities, including those related to gender identity (13), race/ethnicity (68, 69), disabling conditions (70), socio-economic status (71), and immigration status (72).

Although the existence of unequal experiences has been extensively documented and is generally understood by most Americans (73), less widely recognized is an important corollary: that the magnitude and specific patterns of inequities often varies across outcomes in some crucial ways. This was evident in some of our findings. Although those living in low-income households are generally at risk for elevated level of adverse events while receiving healthcare, these risks have in many past studies been concentrated in the lowest quartile of the income distribution. By contrast, findings reported here suggest that the risk of diagnostic mistakes and/or problems is elevated among all households with below-average incomes. Apart from revealing a much wider population at risk, it is these discrepant patterns that offer clues to the origins of certain types of inequitable outcomes.

During the past several decades, patient experience surveys have been widely deployed throughout the U.S. healthcare system, perhaps most impactfully as a means for incentivizing hospitals to promote patient-centered practices (74). Most of these surveys are designed to generate feedback on events that are more prevalent than safety shortfalls, so they have provided relatively little guidance on either the frequency or the inequities in safety experiences, including those occurring during diagnosis, often over time and across multiple settings.

4.5 The role of health delivery systems for the future of diagnostic equity

While the survey results quantify the magnitude of disparities in diagnosis and unveil potential subgroups experiencing diagnostic-related inequities, the implications for the health delivery system may appear hazy. However, when viewed through

the looking glass of potentially different perceptions on the concept of diagnosis—those of diverse patients and clinicians—the need to look anew from all angles merits discussion. When interpreting data derived from patient and care partner experiences, a common critique is that their perceptions about diagnosis may differ from clinical experts, and that the latter somehow trumps the former. Such debates limit subsequent steps to those aimed at sorting out differences between patient and clinician perceptions, as opposed to seizing the opportunity to gain unique insights from diverse and nationally representative samples of the public through surveys such as the one analyzed in this paper. Health delivery systems are in a pivotal position to implement complementary approaches, including stratification of patient level data based on sociodemographic characteristics to evaluate safety and quality disparities. Such stratified analysis would be enriched by expanding patient level data to include questions about experience of the diagnostic process and outcomes. Health delivery system engagement in pursuing incorporation of such data gathering from their patients and neighborhood citizens would facilitate in-depth and local efforts to integrate the complementary expertise of patients, care partners, clinicians and public health officials.

4.6 The role of future research in advancing diagnostic equity

Future research is also pivotal to making progress toward diagnostic equity. First, expanding beyond the illustrative excerpts provided in this paper would include a rigorous qualitative examination of the narratives that accompany the quantitative results from our survey. Second, to the extent that health systems might respond to these results, we anticipate that research that aims to connect diagnostically-focused survey results to currently collected information from health systems about patient satisfaction and experience would be valuable.

4.7 Limitations of the existing study

Despite the valuable insights gained from our study, several limitations must be acknowledged. First, the reliance on self-reported data may introduce social desirability bias, potentially leading to underreporting or overreporting of diagnostic P&Ms and associated outcomes. Past research suggests that patients and care partners will have difficulty separating out diagnostic mistakes that were preventable from adverse events that were not (32). However, understanding both types of diagnostic breakdown is still important and may yield persisting harms, including reducing trust in future diagnostic reliability or safety. Moreover, self-reported outcomes can also identify diagnostic breakdowns that are in clinicians' blindspots, thereby enhancing diagnostic safety (40).

Second, the cross-sectional design of the study precludes establishing causality or temporal relationships between sociodemographic factors and diagnostic outcomes. This limits our ability to infer causation, since statistical associations may embody forms of reverse causality. For example, the odds-ratios identified in the regression models connecting disability status with elevated P&Ms may reflect P&Ms causing work disabilities, rather than patients with disabilities facing greater vulnerability to diagnostic P&Ms.

Third, while efforts were made to ensure the representativeness of the sample to the general U.S. population, inherent biases in survey participation and sampling may have influenced the findings, limiting the generalizability of the results. The most pronounced bias was induced by our reliance on an internet panel for recruiting respondents involves literacy, since people who regularly complete surveys on-line clearly have a reading capacity that is not universal among the American public. That may lead our results to understate the impact of low literacy and limited education on diagnostic outcomes.

Fourth, the use of broad categories for sociodemographic variables based on the questions pre-determined for the nationally representative panel used in this study may overlook the heterogeneity within the available subgroups (e.g., for race and ethnicity, work-related disability status) and obscure important nuances in healthcare experiences and outcomes.

Fifth, while our sample is the largest yet for patient-reported diagnostic P&Ms, it is not large enough for thorough interaction analyses to explore the numerous intersectional predictors worthy of exploration. This is particularly consequential if patients' perception of stigma linked to some personal attribute or aspect of their medical history might become a more pronounced barrier to effective diagnosis for patients who have multiple stigmatizing conditions.

Sixth, in choosing to focus on diagnostic P&Ms from the unique voice and lived experiences of patients and their household care partners, we do not make any claims about clinical adjudication of these reports or potential classifications as diagnostic errors from a medical perspective. Even if diagnostic P&M prevalence and associated harm estimates, along with the sociodemographic patterns revealed in this study, differ to some extent from clinically adjudicated diagnostic errors or breakdowns, this study provides a public health foundation for making progress on diagnostic equity by centering on lived experiences of the public.

Finally, while our study provides valuable insights into demographic disparities in diagnostic outcomes (diagnostic P&Ms and associated harms), the complexity of healthcare disparities warrants further investigation into the underlying mechanisms driving these disparities. Future research employing longitudinal designs and drawing more heavily on qualitative methodologies than did this study may provide a more comprehensive understanding of the factors contributing to diagnostic disparities and inform targeted interventions to improve healthcare equity.

5 Conclusion

In our assessment, this study provides valuable insights into the prevalence and sociodemographic correlates of diagnostic P&Ms, shedding light on the complex interplay between patient

characteristics and healthcare experiences. The findings reveal significant sociodemographic disparities in diagnostic P&Ms, with younger patients, those with lower household income, cis-women, transgender and gender independent individuals, those with individuals with multiracial identities and those who are disabled (unable to have full-time work) being particularly vulnerable.

Moreover, disparities were observed in not only the frequency of diagnostic P&Ms, but also the impact of diagnostic P&Ms, with low-income individuals, cis-females and disabled individuals experiencing higher rates of persistent emotional and physical harm. Younger patients also experience higher rates of persisting emotional harm. These findings underscore the need for targeted interventions to address systemic biases and promote equitable access to high-quality healthcare for all individuals, regardless of their demographic characteristics.

Overall, the findings from this study contribute to a deeper understanding of healthcare disparities and underscore the importance of addressing systemic biases in healthcare delivery. By identifying vulnerable populations and disparities in healthcare experiences, policymakers, healthcare providers, and researchers can develop targeted interventions to improve diagnostic accuracy, enhance patient-provider communication, and promote healthcare equity. Ultimately, addressing demographic disparities in diagnostic P&Ms is essential for achieving the goal of providing high-quality, patient-centered care to all individuals, regardless of their sociodemographic characteristics.

Future research should further explore the underlying mechanisms driving these disparities and evaluate the effectiveness of interventions aimed at mitigating diagnostic P&Ms and errors across diverse sociodemographic groups. By better understanding the origins and implications of disparate diagnostic experiences, we should be able to more effectively identify actionable strategies for reducing the prevalence and impact of diagnostic breakdowns in the future, thereby relieving the burdens on those subgroups who are disproportionately experiencing them now.

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 requirement of ethical approval was waived for the study and deemed exempt by the IRBs at Yale (#2000032012) and Johns Hopkins (IRB00322791) universities. The study fell under the umbrella exemption granted by the University of Wisconsin IRB to qualitative projects conducted by the Qualitative and Health Experiences Research Laboratory in the Department of Family Medicine for the studies involving humans because this research was deemed exempt under 45CFR46.104 (2)(ii). 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 because our study respondents are part of NORC's AmeriSpeak' panel, and consent procedures are reviewed and approved by NORC's IRB.

Author contributions

KM: Conceptualization, Formal analysis, Funding acquisition, Methodology, Visualization, Writing – original draft, Writing – review & editing. KG: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. RG: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. CY: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. ID: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. JE: Conceptualization, Writing – original draft, Writing – review & editing. EW: Writing – original draft, Writing – review & editing. MS: Conceptualization, Formal analysis, Funding acquisition, Methodology, 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.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2025.1444005/full#supplementary-material

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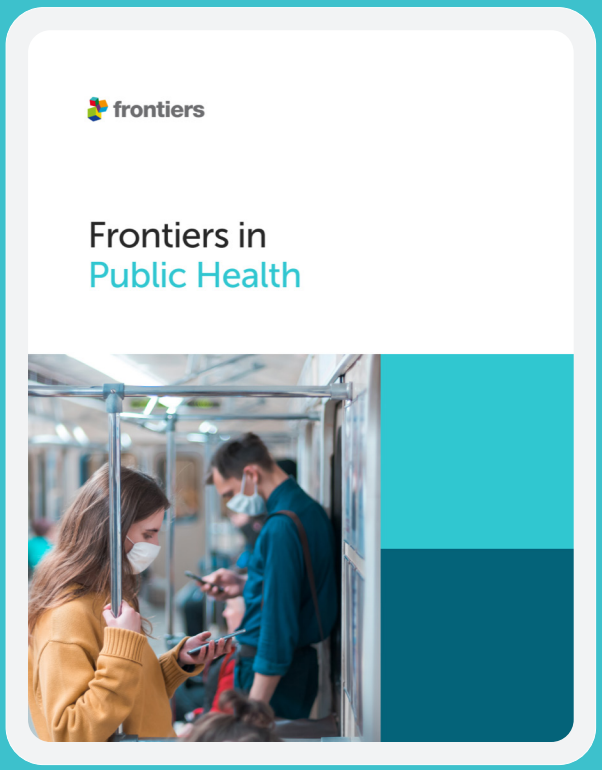

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