Research Topics

World Health Day frontiers in public health presents: “My health, my right”

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Edited by
María Del Carmen Valls Martínez, Barbara Sawicka, Hubert Amu,
Tsitsi Masvawure, Olatunde Aremu, Mo Salman and
Martin Amogre Ayanore

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World Health Day 2024: frontiers in public health presents: “My health, my right”

Topic editors
María Del Carmen Valls Martínez — University of Almeria, Spain
Barbara Sawicka — University of Life Sciences of Lublin, Poland
Hubert Amu — University of Health and Allied Sciences, Ghana
Tsitsi Masvawure — Worcester Polytechnic Institute, United States
Olatunde Aremu — Birmingham City University, United Kingdom
Mo Salman — Colorado State University, United States
Martin Amogre Ayanore — University of Health and Allied Sciences, Ghana

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EDITED AND REVIEWED BY
Xiaozhen Lai,
Peking University, China

*CORRESPONDENCE
Olatunde Aremu
✉ olatunde.aremu@bcu.ac.uk

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Editorial: World Health Day 2024: frontiers in public health presents: "My health, my right"

Olatunde Aremu[1]*, Barbara Sawicka[2], Hubert Amu[3], María Del Carmen Valls Martínez[4] and Tsitsi Masvawure[5]

[1]Department of Life and Sport Sciences, Birmingham City University, Birmingham, United Kingdom, [2]Department of Plant Production Technology and Commodities Science, University of Life Sciences in Lublin, Lublin, Poland, [3]Department of Population and Behavioural Sciences, Fred N. Binka School of Public Health, University of Health and Allied Sciences, Hohoe, Ghana, [4]Mediterranean Research Center on Economics and Sustainable Development (CIMEDES), Economics and Business Department, University of Almería, Almería, Spain, [5]Department of Integrative and Global Studies, Worcester Polytechnic Institute, Worcester, MA, United States

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Editorial on the Research Topic
World Health Day 2024: frontiers in public health presents: "My health, my right"

World Health Day on April 7, 2024, calls for global commitment to the right of individuals to their health with the theme "My health, my right." According to the World Health Organization (WHO), over half the world's population, more than 4.5 billion individuals, did not have access to essential health care, either in terms of physical or mental health, as of 2021 (1). This level of inequality introduces disparities which are shaped by gender, race, education, income, sexual orientation, place of residence, or physical disability (2, 3).

Frontiers in Public Health launched this Research Topic to commemorate the United Nations World Health Day 2024. This Research Topic, bringing together 13 articles, highlights the progress made, the impediments that persist, and the innovative approaches shaping the future of the right to health.

The World Health Organization (WHO) constitution, articulated for the first in 1946, the right to health. Specifically, it states that "the enjoyment of the highest possible standard of health is one of the fundamental rights of every human being irrespective of race, religion, political affiliation, economic status or social condition (4)." This statement by WHO was further enshrined as a human right in 1948 via the Universal Declaration of Human Rights and in 1966 in Article 12 of the International Covenant on Economic, Social and Cultural Rights (ICESCR), which encompasses four core areas—availability, accessibility, acceptability and quality (5). Health is indeed a fundamental human right, and it is important to recognize the need for legal responsibility, fairness, and non-discrimination in its production (5, 6). However, earlier efforts through universal health coverage (UHC), have provided little impetus toward improved healthcare access and impediments have remained in the realization of health for all (6). Although the theme "My health, my right" seems to lend support to an individual's sovereignty, by having a say and unhindered access to information that leads to decisions about their health, there are still challenges to the actualization of such. The advent of emerging and remerging infectious diseases of global health importance, lack of health literacy about availability of services, stigmatization of specific health problems, and cost of healthcare services adversely affect individuals' sovereignty to access health services and their right to health (7–10). Furthermore, the challenges arising from and posed by legislation around personal data privacy from the use of technological innovations have joined forces to make achieving the right to health for individuals somewhat difficult (8, 11).

The articles in this Research Topic address several themes that are relevant to advancing health as a human right. Some articles focus on wider socio-structural factors that impede the right to health, while others propose innovative approaches to ensuring that the right to health is attained for all. In the sections that follow, we offer a summary of the key messages from the articles in the Research Topic and show how they address the theme "My health, my right."

Social determinants of health and health inequalities

Several articles in this Research Topic focus on how social, economic, and environmental factors influence health outcomes and contribute to health inequalities. These articles focus specifically on the high prevalence of non-communicable diseases globally, which is negatively affecting individuals' rights to good health.

In the article by Zhang et al., the authors investigated the relationship between inequalities in chronic respiratory disease (CRD) and all-cause mortality in China and they found that individuals from poor backgrounds had substantially higher risks of chronic lung disease and asthma. This study highlights the need to address the root causes of economic inequalities, and the authors propose interventions that are aimed at improving the educational attainment of individuals from low socioeconomic backgrounds as the key solution.

The article by Liu et al., examines the global epidemiology of pulmonary arterial hypertension (PAH), focusing on trends in incidence, mortality, and disability-adjusted life years (DALYs) over a 32 year period to inform evidence-based policy and healthcare strategies. The findings from the study are novel and reveal an upward trajectory in the incidence of PAH, increasing from 23,301 in 1990 to 43,251 in 2021. Additionally, from 1990 to 2021, PAH-related deaths increased from 14,842 to 22,021. The authors recommend enhanced prevention and comprehensive management strategies as a strategy to shrink the global PAH burden and improve health equity.

The article by Cicekli and Eskin examined the prevalence and co-occurrence of lifestyle risk factors among university students and concluded that targeted interventions, such as promoting physical activity, providing affordable and nutritious meals, and educating students on healthy lifestyles, are essential to reducing non-communicable disease (NCD) risk among students. The study found that students at highest risk of developing NCDs lived in student housing, preferred eating fast foods and watched 4–5 h of television a day. Something about the socio-structural organization of university life seemed to increase students' risk of acquiring NCDs. The article by Cristiane Sibim et al. examined the potential interactions between socioeconomic indicators and the One Health Index (OHI) in South American countries and the authors found, surprisingly, that better environmental health was not associated with better human or animal health. The authors, as part of their findings, considered absence of stronger indicators for animal health to be a key gap in better understanding the interplay of One Health factors. A key finding the authors also highlight is that social factors, rather than economic factors like GDP, seem to explain differences in the One Health status observed in South American countries.

The last article we will highlight in this section is by Bezie et al. who investigated the prevalence of work-related burnout and its correlates among kindergarten teachers in Dessie City, Northeast Ethiopia. Results from bivariate logistic regression and multivariable regression models revealed the total prevalence of Work-related burnout (WRB) was 39.7% [95% CI (34.8, 44.6)]. Work–family conflicts, working conditions, perceived stress, job dissatisfaction, and kindergarten type were all significantly associated with WRB. Some of the social determinants of work-related burnout included long work hours and a lack of appreciation of the emotionally draining nature of kindergarten care. The authors recommend interventions focused on improving school environments to be more supportive of the wellbeing of teachers.

Innovative approaches and interventions to address health disparities

Several articles in this Research Topic focus on the application of novel approaches, interventions, and best practices that can help combat health disparities, improve health equity, and safeguard the health rights of diverse people. The article by Sapp et al. discusses the development, implementation and evaluation of a pilot of two medical education interventions aimed at improving attitudes and empathy toward individuals with disability among medical students in the U.S. The findings show that one of the interventions, the elective course, but not the 2 hr course, significantly decreased student anxiety levels, likely due to more individual time working with individuals with disabilities. However, delayed analysis after 3 months showed that both interventions had a lasting impact on attitudes and behavior change when caring for individuals with disabilities. This study draws attention to the growing population of individuals with disabilities in the U.S. and the importance of ensuring that healthcare providers are well-trained to provide high quality services to this population.

The article by Chen and Tian assessed the financial effectiveness of a combination of modified gemcitabine and oxaliplatin in the management of gall bladder cancer (GBC) in China, using data from a randomized controlled study in individuals with metastatic GBC. The cost-effectiveness analysis concludes that in a Chinese healthcare context, modified gemcitabine coupled with oxaliplatin (mGEMOX) is not a cost-effective treatment option for unresectable GBC. This paper is important for highlighting the costs of medical treatments as key impediments to the realization of health for all.

Similarly, the article by Schenkman and Bousquat examined efficiency and effectiveness in health services in remote rural localities (RRL) compared to urban and rural communities of Brazil. The authors report that Brazilian RRL localities show superior resource and health efficiency largely due to how primary healthcare teams are organized. The authors concluded that reducing intersectional inequities in income and education by ethnicity could greatly increase the efficient attainment of health levels in society. This study is innovative in its attempt to quantify health provision and outcomes in the most remote of rural areas, which tend to be hard to reach and often not consistently included in national-level statistics. Remote rural communities often constitute some of the most invisible and vulnerable populations globally.

Disparities in equity in access to healthcare services and outcomes among diverse populations

The importance of equity in access to, and utilization of healthcare services in improving the wellbeing of diverse populations cannot be overemphasized. In the article by Bu et al. the authors explored the heterogeneity of public health service use and how it relates to the social integration of older adult migrants in China. The authors concluded that many aspects influence utilization of public health services amongst older adult migrants, such as gender, education and extent of mobility. They also found that familiarity with local resources increased public health service utilization. Thus, the authors recommend more targeted policies that are user-friendly to help improve uptake of public health services for older adult migrants.

Community empowerment and participation

The importance of initiatives that empower communities to advocate for their health rights, participate in decision-making processes, and contribute to improving health outcomes cannot be overstated. In the article by Borondy-Jenkins et al., the Hepatitis B Foundation (HBF) convened the first global hepatitis B and hepatitis delta Community Advisory Board (CAB) with 23 members from 17 countries, representing six out of the seven World Health Organization (WHO) regions, and countries with the largest hepatitis B and hepatitis delta disease burden. The aim was to reflect on the process of assembling an effective and motivated CAB and assess the impact on CAB participants. Three virtual focus group sessions were held with 16 participants in July and August 2023. Participants reported that through CAB membership, they gained networking and advocacy opportunities, as well as enhanced their knowledge of hepatitis B and hepatitis delta. The authors recommend that a regular internal review of the community advisory boards' structure and performance is critical to ensure the CAB is fulfilling its mission.

Health policy and governance

Periodic assessment of the effectiveness of policies, strategies, and governance structures in promoting health equity and upholding the rights of individuals to health is essential. The article by Külper-Schiek et al. reported the outcome of an evaluation of the effectiveness of national immunization technical advisory groups (NITAGs) in middle-income countries funded by the WHO Regional Office for Europe and the Robert Koch Institute (RKI). The findings show that all the NITAGs studied lacked a well-staffed Secretariat to establish annual work plans and develop NITAG recommendations following a standardized process. The authors recommend that the WHO and NITAG partners continue to provide training on the standardized recommendation-making process and advocate for increased MoH support to NITAGs, including dedicated Secretariat staff. NITAGs play a critical role in ensuring easy access to life-saving immunizations and are thus key partners in the quest for health justice.

Ethical dilemmas and considerations

Addressing ethical dilemmas that may arise in the provision of healthcare services is crucial to ensuring that individual rights are respected, and equitable health outcomes are maintained. The article by El Bouchikhi et al. is a scoping review that explores opportunities and ethical issues that are inherent in the use of digital technologies and the Internet of Things (IoT) within occupational safety and health (OSH). The review identifies many ethical issues but notes that these provide key information and guidance for those who wish to develop evaluation frameworks in line with a preventive regulatory approach. More importantly, the list informs policymakers and practitioners about the governance of such tools for ensuring more OSH.

Conclusion: moving toward achieving the right to health

We, the Editors, strongly believe that the theme "My health, my right" is a call to renew action toward empowering individuals to achieve the needed right to health. The selection of articles from various regions globally present the current situation regarding the right to health from various dimensions.

Ultimately, to achieve the WHO declaration of equal rights to health and enhance individuals' long-term health and wellbeing, deep-rooted systemic injustice in access to healthcare, which manifests in several ways, must be fully eliminated (7, 12). Unparalleled and sustainable commitment to tackle disparities in access to and utilization of healthcare services for vulnerable groups must be pursued (3, 13). Health system strengthening is imperative and needed to ensure health equity; however, the need for multisectoral and community participation in spreading health awareness is essential and should be widely promoted (14). Participation requires enabling health service users, groups and civil society to engage in planning, decision-making, and implementation processes for health across all levels of the system

(15). Literacy is vital in assimilation and comprehension of health information; hence, investment in such should be accorded a greater priority (16).

Advancements in the deployment of digital technologies have brought new hope in ensuring that people can access healthcare without geographical boundaries (17). Despite this, inequities persist along the lines of income, education, geography, and gender, disproportionately affecting vulnerable populations (2, 9, 10). The UHC, rooted in primary health care, can help countries accomplish the right to health by making sure all people have affordable, equitable access to health services (6). Notwithstanding all the efforts being made, it is ultimately the responsibility of individuals to take control of their health.

Author contributions

OA: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. BS: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – review & editing. HA: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – review & editing. MV: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – review & editing. TM: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – review & editing.

Conflict of interest

The author declares that the research was conducted without commercial or financial relationships that could create a conflict of interest.

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Heterogeneity in public health service utilization and its relationship with social integration among older adult migrants in China: a latent class analysis

Xiaolong Bu, Ting Wang, Qian Dong and Cuiping Liu*

School of Nursing, Shandong First Medical University, Tai'an, China

Background: The older adult migrant population in China is on the rise, which presents challenges for the national public health service system. However, the heterogeneity of public health service utilization and its relationship with social integration among the older adult migrant population remains unclear. This study aims to explore the heterogeneity the public health service utilization and how it relates to their social integration.

Methods: A total of 6,178 older adult migrants from the China Migrants Dynamic Survey (CMDS) in 2017 were included in this study. Exploratory factor analysis was used to categorize social integration into four dimensions. Latent class analysis (LCA) was used to identify different sub-groups of public health service utilization. ANOVA and multivariate logistic regression were used to determine the characteristics of different sub-groups.

Results: Three potential classes of public health service utilization were identified: low utilization of basic public health services class ($N = 3{,}264{,}52.756\%$), medium utilization of basic public health services class ($N = 1{,}743{,}28.172\%$), and high utilization of basic public health services class ($N = 1{,}180{,}19.072\%$). Gender, education, extent of mobility, and move alone or not, flow time were all predictors of the class of public health service utilization. There were significant differences in social integration across potential categories ($p<0.0001$).

Conclusion: The utilization of public health services of the older adult migrants is affected by many aspects. Social integration deserves attention as a significant influencing factor in the utilization of public health services. The government should pay attention to the characteristics of the older adult migrants and formulate relevant policies in a targeted manner in order to improve the utilization of public health services of the older adult migrants.

KEYWORDS

exploratory factor analysis, latent class analysis (LCA), older migrant population, social integration, utilization of basic public health services

EDITED BY
Mo Salman,
Colorado State University, United States

REVIEWED BY
Shanlong Ding,
World Health Organization, Switzerland
Heng Piao,
Affiliated Cancer Hospital of Zhengzhou University, China
Noshin Farzana,
International Center for Diarrhoeal Disease Research (ICDDR), Bangladesh

*CORRESPONDENCE
Cuiping Liu
✉ lcping0926@126.com

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1 Introduction

As China progresses through industrialization and socialist modernization, the number of floating migrants is rapidly increasing. Recent data from the seventh national census indicates that the floating population in China has surpassed 370 million, indicating a 69.73% surge since the sixth national census in 2010 (1). The increasing floating population is crucial for the economic and social progress of the nation. With the rapid development of the economy and society, China is experiencing a rapid aging of its population and changes in demographic composition. As of the end of 2021, the population aged 60 and above had reached 267 million, representing 18.9% of the total population (1). Correspondingly, the number and proportion of older adult individuals within the floating population have also surged. According to data from the 2018 national dynamic monitoring of the floating population, older adult individuals comprised 39.7% of the floating population, including 60,000 individuals in the older age group (2). The increase in older adult migrants moving between regions has given rise to a range of societal challenges such as health issues (3), social isolation (4), concerns regarding psychological well-being (5, 6), and obstacles related to social welfare and integration (7). Research indicates that older adult members of the floating population are more susceptible to health problems and have a lower quality of life compared to younger migrants or local senior citizens (8, 9).

Providing public health services to its citizens has long been a persistent policy program in China (10). In 2009, China launched a new health reform endeavor, introducing the national basic public health service system (NBPHS) plan under the auspices of the health ministry (11). The primary objective of this initiative was to provide complimentary and discretionary basic health services, covering maternal and infant health, mental health management, older adult health management, and areas such as tuberculosis. Studies have underscored the significant impact of public health services on the health outcomes of older adult migrants. The government offers services such as physical examinations and health education to the floating population, facilitates access to medical care for the older adult floating population, and establishes health records through public health service projects. Research indicates that these interventions can positively influence the self-perceived health status and management of chronic conditions among the older adult floating population (12–15).

Various factors can influence the utilization of public health services among the older adult migrant population. Current research demonstrates that factors such as age, gender, and general demographic characteristics may differ among older adult migrants who access public health services (16, 17). Additionally, social integration can play a pivotal role. Social integration, a concept initially introduced by French scholar Émile Durkheim, posits that strong social integration can mitigate the incidence of suicide. Subsequently, scholars have further explored and refined this concept. Chinese scholar Yuan (18) defined social integration as the establishment of connections among diverse cultures, groups, and individuals based on mutual familiarity and adaptation. This process of establishing connections is crucial for promoting social cohesion. Concerning the social integration of the floating population, Sun (19) sees social integration as the process by which immigrants grow into local members and are empowered to participate in various social relationships. Given their limited adaptability and cognitive abilities (20), the challenge of social integration is particularly acute among older adult migrants compared to other cohorts. Effective integration can help older adult migrants in acclimating to their new environment, enhancing their involvement in local social activities, fostering better interpersonal communication, and ultimately elevating their quality of life and well-being. The struggle of older adult migrants to integrate seamlessly into their new surroundings can impede their access to public health services. However, the precise impact of social integration on the utilization of public health services among older adult migrants remains uncertain.

Limited research currently exists on the relationship between social integration and the utilization of public health services among migrants. Existing studies have primarily focused on demographic factors influencing the use of public health services. For instance, Yanwei Lin and colleagues examined public health service utilization among the older adult floating population, with a focus on age and medical insurance as key factors influencing their access to these services (21). Similarly, Dan Tang et al. explored the utilization of public health services among the older adult floating population, highlighting demographic factors such as household registration type, income level, and education in determining their engagement with these services (11). XueYao Wang investigated the impact of social integration on migrant populations' access to health education and healthcare choices in a cross-sectional survey (22), representing the sole study known to explore the link between public health service utilization and social integration among migrants. Nevertheless, this research does not account for individual differences in service utilization or how the social integration of migrants across various age groups may influence their use of public health services. Other studies on public health service utilization by migrants have overlooked the heterogeneity within this population. Latent class analysis (LCA), a person-centered approach, can identify potential subgroups based on individual responses, thereby categorizing the population into several categories (23). LCA proves suitable for this study as the utilization of public health services among the older adult migrant population is inevitably heterogeneous due to the differences in educational level, household registration status, mobility patterns, and various other factors.

The objective of this study was to differentiate individual differences in the utilization of public health services among the older adult migrant population using LCA. Additionally, the study was also aimed to investigate how demographic characteristics and social integration impact their engagement with public health services.

2 Methods

2.1 Data source

The data used in this study was obtained from the 2017 China Migrants Dynamic Survey (CMDS), which employed a rigorous sampling method to gather information from 169,989 individuals within the floating population aged 15 and above. These individuals had been residing locally for at least 1 month and did not possess household registration in the area. According to the National Standard for New Basic Public Health Service (NBPHS) (third edition), the

TABLE 1 Evaluation indicators of social integration.

Item	Evaluation
What is your average monthly income in the past year?	0 ~ 1,999 = 1, 2,000 ~ 3,999 = 2, 4,000 ~ 5,999 = 3, Above the 6,000 = 4
What is your average monthly expenditure in the past year?	0 ~ 1,999 = 1, 2,000 ~ 3,999 = 2, 4,000 ~ 5,999 = 3, Above the 6,000 = 4
I like the city	1 = completely disagree; 2 = disagree; 3 = basic agree; 4 = complete agree
I noticed the changes in this city	1 = completely disagree; 2 = disagree; 3 = basic agree; 4 = complete agree
I would like to integrate into the local people and become one of them	1 = completely disagree; 2 = disagree; 3 = basic agree; 4 = complete agree
Since 2016, have you made advice or supervised the unit / community / village management	1 = no; 2 = occasionally; 3 = sometimes; 4 = often
Since 2016, have you reported the situation to relevant government departments in various ways	1 = no; 2 = occasionally; 3 = sometimes; 4 = often
My personal hygiene habits are not different from those of local citizens	1 = completely disagree; 2 = disagree; 3 = basic agree; 4 = complete agree
Following the customs of my hometown is not important to me	1 = completely disagree; 2 = disagree; 3 = basic agree; 4 = complete agree

initial step to access public health services in one's place of residence is to establish a health record. To fulfill this requirement, individuals must have resided in their current location for more than 6 months. Hence, this study specifically focused on mobile older adult individuals aged over 60 who met this residency criterion. After excluding non-compliant and invalid data, a total of 6,178 subjects were included in the analysis.

2.2 Variables

2.2.1 Dependent variables

The National Health and Family Planning Commission launched a pilot program for equal access to NEPHS for migrants in 2013 and issued Guidance on the management of basic public health services for migrants in 2014. The guidelines identified six most important public health services for migrants as childhood vaccines, prevention and control of communicable diseases, maternal and child health, health records, family planning and health education (24). In the China Migrants Dynamic Survey, there was a section that focused on the utilization of public health services. Two aspects including ten items that are most pertinent to older individuals were chosen to evaluate their utilization of public health services in our study. These items have been previously validated in similar studies (11, 25). The first aspect concentrated on the establishment of health records and included one item. The second aspect pertained to the acceptance of health education and encompassed nine items: occupational diseases, tuberculosis, chronic diseases, prevention of sexually transmitted diseases, control of smoking, maternal and child health care, reproductive health, mental health, and emergency response to public emergencies. This ten items are binary variables that required a "yes" or "no" response. For the purpose of latent class analysis, we consider the "yes" option as "1" and the "no" option as "0".

2.2.2 Independent variables

The study focused on the social integration of the older adult floating population as the main independent variables. After conducting a thorough literature review and consulting with the research group, a set of nine items was selected to evaluate the social integration of this population (26). To facilitate subsequent analysis, an exploratory factor analysis was performed to process and analyze the data. The specific items and their corresponding scores are shown in Table 1.

2.2.3 Covariates

In recent years, scholars have focused on the utilization of public health services by migrants. Extensive research has demonstrated that sociodemographic characteristics such as age, gender, educational status, marital status, and household registration are linked to the utilization of health services. The household registration policy, enacted by the Chinese authorities in 1958, divides household registration by provinces and is categorized as either rural or urban. The household registration system allows individuals to receive social benefits in the area of their household registration, but also restricts access to social benefits outside the household registration. Individuals' household registration is not fixed, as they can obtain a household registration in their place of residence through work or education.

Additionally, factors related to migration are also believed to influence the usage of public health services (22). Drawing upon existing studies and questionnaires, we chose six sociodemographic characteristics and four migration-related characteristics as covariates. The sociodemographic characteristics included gender (female or male), age (60–70, 70–80, above 80), education (primary school and below, junior middle school, senior middle school, college degree or above), household registration (rural or urban), marital status (married or single), and health condition (poor health or good health). The migration-related characteristics include whether the individual migrated alone (yes or no), whether they have settled down (yes or no), and the scope of migration (inter-provincial, intra-provincial, or intra-city across counties), flow time(less than 1 year, 2–5 years, 6–9 years, more than 10 years).

2.3 Statistical analysis

Latent class analysis(LCA) was performed using Mplus7.4. LCA is a method used to account for the diversity within a population in observational data by identifying potential subgroups of individuals. This allows for the examination of how older adult migrants utilize public health services based on different characteristics within the population. The most suitable model is chosen by assessing various fit indices, such as the Akaike information criterion (AIC), Bayesian information criterion (BIC), sample size adjusted BIC (SSA-BIC), Lo–Mendell–Rubin likelihood ratio test (LMR-LRT), and bootstrap likelihood ratio test (BLRT). Lower values of AIC, BIC, and SSA-BIC indicate a better fit, with BIC being the most reliable criterion. The significance of p-values for LMR-LRT and BLRT results is important ($p < 0.05$) in determining the validity of the model. Additionally, the interpretability of the model should also be taken into account.

Data were analyzed using SPSS 21. The dimensions of social integration were analyzed using exploratory factor analysis (27). Categorical variables were described with frequencies and percentages. Continuous variables were presented with means with standard deviations (SD). Chi-square tests were used to examine between-group differences for categorical variables, while ANOVA was used for the comparison among continuous variables, followed by *post hoc* analyses, and the significance levels were adjusted using the Tukey HSD method. Finally, multivariate logistic regression was used to investigate the factors influencing the potential categories of public health service utilization among the older mobile population.

3 Results

3.1 Basic information of the older adult floating population

The study consisted of 6,187 older adult migrants, with an average age of 66.09 ± 5.62 years. The majority of participants (77.8%) fell within the 60 to 70 age range. Out of the total sample, 3,581 participants were male (57.9%), while 2,606 were female (42.1%). A significant portion (47.8%) of the participants had completed only primary school or below, amounting to 2,955 individuals. Furthermore, 3,553 participants held a rural household registration, accounting for 57.4% of the population. The majority of participants (84.1%) were married, amounting to 5,204 individuals, while 1,497 individuals were solo migrants (24.2%). Approximately 44% of the older adult floating population expressed a desire to settle in their current location. In terms of self-reported health status, the majority (81.2%) described themselves as healthy or fairly healthy. Regarding migration patterns, 2,734 participants migrated between provinces (44.2%), while 34.8% moved within cities and 21% moved between counties. Only 9% of older adult migrants have been flown for less than a year. Totaling 2,339 older adult migrants, have been flown for over 10 years, making up 37.8% of the total. Descriptive statistics for these variables are presented in Table 2.

3.2 Results of LCA of public health services

As shown in Table 3, this study adopted five models to conduct LCA. The AIC and BIC values showed a gradual decrease across the models, whereas both LRT and BLRT values were found to be statistically significant in all five models. Additionally, the entropy surpassed 0.8 for each of the models. Based on the proportions of different categories in the table, it becomes apparent that if there are more than three categories and the potential categories account for less than 10%, a substantial disparity would exist between the samples. It has the potential to introduce inaccuracies into the model. Therefore, considering the actual situation and the accuracy of the results, it is advisable to opt for the most concise model, namely the third model, which classifies the utilization of public health services by the older adult floating population into three potential categories.

Class 1, named "low utilization of basic public health services class," had the largest sample size of 3,264 cases (52.756%). The majority of subjects in this class did not have established health records and did not receive any health education. However, a few individuals had received education on smoking cessation and the prevention and treatment of chronic diseases. Class 2, named "medium utilization of basic public health services class," consisted of 1,743 patients (28.172%). This class had intermediate levels of established health records and received some health education. Class 3, named "high utilization of basic public health services class," had the smallest sample size of 1,180 cases (19.072%). This class had the best-established health records and the highest level of health education. These classes were identified based on the patterns observed in the data, and their distribution can be visualized in Figure 1.

3.3 Differences of social integration in the utilization of public health services

KMO and Bartlett's test of sphericity for the selected indicators of social integration showed that they were suitable for factor analysis (KMO = 0.63; Bartlett's test of sphericity $\chi^2 = 13391.969$, $p < 0.001$). Based on the outcomes of the exploratory factor analysis, four common factors were derived and designated as psychological integration, economic integration, social participation integration, and cultural integration (Table 4). Psychological integration among older adult migrants refers to their inner thoughts and feelings about integrating into their new place of residence. This can be measured by their expressions of affection and concern for the new location, as well as their subjective willingness to become a part of the local community. Economic integration is assessed through indicators such as monthly income and expenditure, providing insight into the financial situation and spending habits of older adult migrants. Social participation integration examines the extent to which older adult migrants engage in social activities within their new communities, focusing on their involvement in community management and provision of policy advice to the government. Cultural integration assesses the degree to which older adult migrants have embraced the cultural norms and practices of their new environment, particularly in terms of personal hygiene and attitudes towards local customs. The scores corresponding to each common factor, as well as the total score for social integration, were computed based on the assigned scores. A higher score reflected a better overall level of social integration. The older adult floating population showed an average social integration score of 22.04 ± 2.741, with the highest score of 3.49 ± 0.514 in psychological integration. It was followed by economic integration with the score of 2.42 ± 0.929, cultural integration with the score of 2.26 ± 0.688, and the lowest score of 1.10 ± 0.342 was observed in the social participation integration dimension. The ANOVA results indicated a significant difference in social integration levels among the three potential classes of public health service utilization. *Post hoc* comparisons revealed that the class with high utilization of public health services obtained notably higher scores in psychological

TABLE 2 Public health service subgroups with different demographic characteristics.

Characteristics		Overall situation	Class 1 (*n* = 3,264)	Class 2 (*n* = 1,743)	Class 3 (*n* = 1,180)	χ^2	p
Gender	Male	3,581 (57.9%)	1,833 (56.2%)	1,039 (59.6%)	709 (60.1%)	8.462	0.015
	Female	2,606 (42.1%)	1,431 (43.8%)	704 (40.4%)	471 (39.9%)		
Age	60–70	4,814 (77.8%)	2,503 (76.7%)	1,358 (77.9%)	953 (80.8%)	16.146	0.003
	70–80	1,173 (19.0%)	638 (19.5%)	327 (18.8%)	208 (17.6%)		
	Above 80	200 (3.2%)	123 (3.8%)	58 (3.3%)	19 (1.6%)		
Education	Primary school and below	2,955 (47.8%)	1,635 (50.1%)	843 (48.4%)	477 (40.4%)	41.642	0.000
	Junior middle school	1,867 (30.2%)	934 (28.6%)	550 (31.6%)	383 (32.5%)		
	Senior middle school	967 (15.6%)	490 (15.0%)	255 (14.6%)	222 (18.8%)		
	College degree or above	398 (6.4%)	205 (6.3%)	95 (5.5%)	98 (8.3%)		
Household registration	Rural	3,553 (57.4%)	1939 (59.4%)	1,013 (58.1%)	601 (50.9%)	25.927	0.000
	Urban	2,634 (42.6%)	1,325 (40.6%)	730 (41.9%)	579 (49.1%)		
Marital status	Married	5,204 (84.1%)	2,722 (83.4%)	1,472 (84.5%)	1,010 (85.6%)	3.345	0.188
	Single	983 (15.9%)	542 (16.6%)	271 (15.5%)	170 (14.4%)		
Whether to flow alone	Yes	1,497 (24.2%)	757 (23.2%)	421 (24.2%)	319 (27.0%)	6.975	0.003
	No	4,690 (75.8%)	2,507 (76.8%)	1,322 (75.8%)	861 (73.0%)		
Whether to settle down	Yes	2,722 (44.0%)	1,449 (44.4%)	736 (42.2%)	537 (45.5%)	3.952	0.413
	No	2,249 (36.4%)	1,186 (36.3%)	652 (37.4%)	411 (34.8%)		
	Not clear	1,216 (19.7%)	629 (19.3%)	355 (20.4%)	232 (19.7%)		
Health condition	Health	5,021 (81.2%)	2,568 (78.7%)	1,435 (82.3%)	1,018 (86.3%)	34.877	0.000
	Ill health	1,166 (18.8%)	696 (21.2%)	308 (17.7%)	162 (13.7%)		
Flow range	Trans-provincial	2,734 (44.2%)	1,558 (47.7%)	706 (40.5%)	470 (39.8%)	39.645	0.000
	Inter-city within the province	2,156 (34.8%)	1,092 (33.5%)	640 (36.7%)	424 (35.9%)		
	Within the city across the county	1,297 (21.0%)	614 (18.8%)	397 (22.8%)	286 (24.2%)		
Flow time (year)	≤1	559 (9.0%)	301 (9.2%)	164 (9.4%)	94 (8.0%)	15.573	0.016
	2–5	1903 (30.8%)	988 (30.3%)	528 (30.3%)	387 (32.8%)		
	6–9	1,386 (22.4%)	699 (21.4%)	389 (22.3%)	298 (25.3%)		
	≥10	2,339 (37.8%)	1,276 (39.1%)	662 (38.0%)	401 (34.0%)		

integration and social participation compared to those in class 1 and class 2. Additionally, class 2 scored higher than class 1 in these domains. In terms of economic integration, although no significant difference was found between class 1 and class 2, class 3 scored significantly higher than both. Class 1and class 2 had significantly higher scores in cultural integration compared to class 3. Specific scores are shown in Table 5.

3.4 Results of multivariate logistic regression

When examining the differences between class 1 and class 2 within the older migrant population, several findings emerged. Firstly, it was found that males (OR = 1.134, $p = 0.001$) and those with lower secondary education (OR = 1.332, $p = 0.002$) were more likely to belong to class 2. Conversely, migrants who moved across provinces were more inclined to be classified in class 1 (OR = 0.705, $p < 0.001$). Additionally, individuals with better psychological and social integration were more likely to be categorized into class 2 (OR = 1.222, $p = 0.001$; OR = 1.761, $p < 0.001$). In comparing class 1 and class 3, it was found that those who migrated alone were more likely to belong to class 3 (OR = 1.218, $p < 0.001$), while those who moved across provinces or cities were more likely to be classified in class 1 (OR = 0.582, $p < 0.001$; OR = 0.804, $p < 0.001$). Social integration also played a significant role in the categorization of class 1 and class 3. Individuals with better psychological, social participation, and economic integration had a higher likelihood of belonging to class 3

TABLE 3 Model fit statistics for latent class analysis models specifying one to five classes.

Model	AIC	BIC	SSA-BIC	Entropy	LMR-LRT	BLRT	Class size (%)
1	74597.3	74664.6	74632.8	–	–	–	–
2	52676.4	52817.8	52751.0	0.932	<0.00001	<0.00001	31.6/68.4
3	**49443.5**	**49658.9**	**49557.2**	**0.876**	**<0.00001**	**<0.00001**	**52.8/28.2/19**
4	49012.8	49302.2	49165.6	0.880	<0.00001	<0.00001	22/18.6/6.4/53
5	48600.3	48963.7	48792.1	0.830	<0.00001	<0.00001	7.4/16.4/19.3/7/50

AIC, Akaike Information Criteria; BIC, Bayesian Information Criteria; SSA-BIC, Adjusted Bayesian Information Criteria; LMR-LRT, LoMendell-Rubin Likelihood Ratio; BLRT, Bootstrapped Likelihood Ratio Tests. Smaller numbers for AIC, BIC, and SSA-BIC metrics represent a better model fit.
The *p*-value of LMR-LRT, BLRT is significant then it means that the fit of model K is better than that of model K−1. Bolded row represents the selected model.

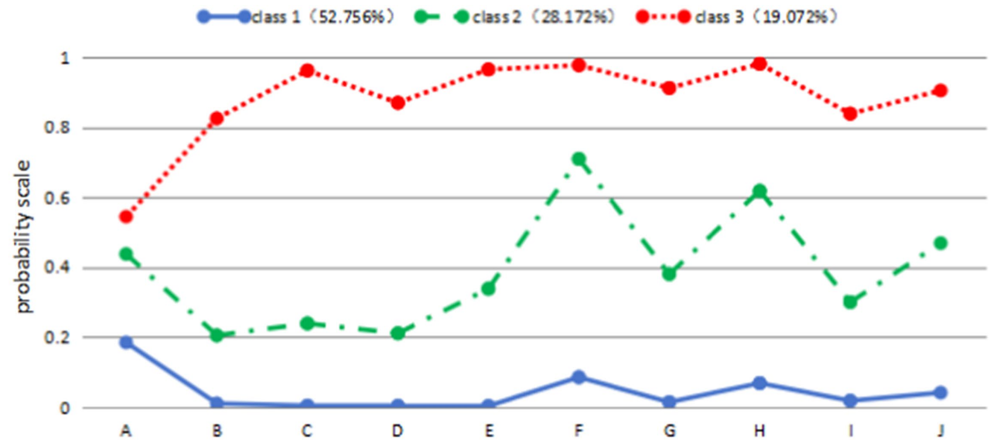

FIGURE 1
Distribution of characteristics of three potential subgroups of public health service utilization among older mobile populations. (A) Health records; (B) occupational disease; (C) sexually transmitted diseases; (D) reproductive health; (E) tuberculosis control; (F) smoking education; (G) mental health; (H) chronic disease; (I) maternal and child health; (J) public emergencies.

TABLE 4 Post-rotation social integration factor components.

Item	Factor			
	Psychological integration	Economic integration	Social participation integration	Cultural integration
The degree of inflow of liking	**0.871**	0.004	0.008	−0.079
The degree of inflow of concern	**0.858**	0.015	0.046	−0.042
The willingness to merge into the ground	**0.848**	0.004	0.014	−0.092
Monthly income	−0.012	**0.913**	0.047	−0.025
Monthly expenditure	0.030	**0.912**	0.029	−0.32
Willingness to participate in public affairs	0.023	0.021	**0.867**	0.012
Provide policy advice	0.031	0.052	**0.864**	−0.024
Health habit	0.000	0.028	−0.019	**0.831**
Social customs	−0.169	−0.082	0.008	**0.763**
Eigenvalues	2.244	1.674	1.504	1.291
% of variance explained	24.933	18.597	16.712	14.345
% of total variance explained	74.588			

Bold values in the same column indicate that the items are categorized in the same factor.

(OR = 1.685, $p < 0.001$; OR = 2.425, $p < 0.001$; OR = 1.086, $p = 0.028$), whereas those with better cultural integration were more likely to be excluded from class 3 (OR = 0.841, $p < 0.001$). Lastly, when comparing class 2 and class 3, individuals with better psychological, social participation, and economic integration were more likely to belong to class 3 rather than class 2 (OR = 1.369, $p < 0.001$; OR = 1.326, $p = 0.002$; OR = 1.092, $p = 0.046$). Detailed data are shown in the Table 6.

TABLE 5 Social integration score of the older mobile population and implications for public health service subgroup.

	Class 1 (*n* = 3,264)	Class 2 (*n* = 1,743)	Class 3 (*n* = 1,180)	F	*p*	Tukey HSD
Psychological integration	3.44 ± 0.516	3.51 ± 0.504	3.60 ± 0.507	38.774	0.000	1<2 2<3 1<3
Economic integration	2.40 ± 0.951	2.39 ± 0.923	2.51 ± 0.864	7.120	0.001	1<3 2<3
Social participation integration	1.06 ± 0.272	1.12 ± 0.361	1.18 ± 0.453	55.582	0.000	1<2 2<3 1<3
Cultural integration	2.30 ± 0.693	2.26 ± 0.665	2.18 ± 0.699	6.552	0.000	3<1 3<2
Total score	21.86 ± 2.735	22.04 ± 2.725	22.52 ± 2.724	189.563	0.000	1<3 2<3

TABLE 6 Multiple logistic regression analysis of public health service utilization among the older adult mobile population.

	β	OR	95%CI	*p*
Class 2 vs. Class 1 (class 1 as a reference class)				
Gender: male	0.126	1.134	1.002–1.284	0.001
Education: junior high school	0.287	1.332	1.008–1.761	0.002
Flow range: across provinces	−0.350	0.705	0.601–0.827	0.000
Psychological integration	0.201	1.222	1.089–1.372	0.001
Social participation integration	0.566	1.761	1.455–2.133	0.000
Class 3 vs. class 1 (class 1 as a reference class)				
Flow alone: Yes	0.197	1.218	1.035–1.432	0.000
Flow range: across provinces	−0.541	0.582	0.484–0.700	0.000
Flow range: cross-city	−0.218	0.804	0.668–0.968	0.000
Psychological integration	0.522	1.685	1.464–1.939	0.000
Social participation integration	0.886	2.425	2.004–2.935	0.000
Economic integration	0.082	1.086	1.009–1.168	0.028
Cultural integration	−0.173	0.841	0.762–0.929	0.000
Class 3 vs. class 2 (class 2 as a reference class)				
Psychological integration	0.314	1.369	1.172–1.599	0.000
Social participation integration	0.282	1.326	1.108–1.586	0.002
Economic integration	0.008	1.092	1.001–1.191	0.046

4 Discussion

This study explored the connection between public health service utilization and social integration among the older adult migrant population, utilizing data from the 2017 CMDS. The results of LCA indicated that most of the population had inadequate access to public health services, categorized as class 1 and class 2. Less than 20 % belonged to class 3. These results underscore the suboptimal utilization of public health services among the older adult population (28). Specifically, class 1 demonstrated poor overall utilization of public health services, excluding health record establishment and education on smoking and chronic diseases. This could be attributed to their limited health awareness and hesitance to participate in health education and public health services. Their willingness to engage in education on chronic diseases and smoking, however, suggests a selective acceptance of health education within this class. In comparison, class 2 showed a moderate level of acceptance towards public health services compared to the other two classes. They possessed sound knowledge of public health services, demonstrated high health awareness, and actively engaged in such services. However, they showed less involvement in the prevention and treatment of occupational and sexually transmitted diseases, as well as reproductive health and maternal and child care. They had a preference for health education on smoking and chronic diseases. Additionally, both class 1 and class 2 displayed a higher demand for knowledge regarding smoking and chronic diseases. Therefore, it is recommended that relevant departments provide targeted health education to address the specific needs of the older adult migrant population. On the other hand, class 3 demonstrated the highest level of engagement in public health services, exhibiting a strong interest in various health education programs. Although their participation in health records was not significantly different from class 1 and class 2, this can be attributed to government policies and oversight in this domain. Acceptance of health education is largely driven by individual willingness, with those who are more health-conscious being more likely to engage in such programs. In contrast, the establishment of health records is overseen by government and healthcare institutions, making individual health awareness less influential in this regard.

The complex nature of social integration among the floating population can be examined from various perspectives. In this study, social integration was specifically divided into four dimensions: psychological integration (level of attachment, level of concern, willingness to assimilate), economic integration (income, expenses), social participation integration (willingness to engage in public affairs, providing policy recommendations), and cultural integration (health practices, societal norms). The results demonstrated that the older adult floating population displayed the highest levels of psychological integration, indicating a strong emotional connection and a desire to adapt to their current location. However, in addition to social participation integration, which scored the lowest, older people's cultural integration scores were also at the lower end of the scale. This suggests difficulties in embracing the customs, traditions, and health behaviors of their new environment. On one hand, their aspiration for a safe and happy life drove their strong inclination towards psychological integration. On the other hand, their traditional values of honoring the older adult and cherishing their hometown influence their adherence to the customs and cultural norms of their hometown even in their new residence (29, 30).

Based on the findings regarding the impact of social integration on the utilization of public health services, it could be concluded that each dimension significantly influenced the utilization of such services. Notably, improved cultural integration among the older adult floating population was associated with a decreased utilization of public health services, unlike the other dimensions. This association might be attributed to educational background, which exerted a significant impact on both the cultural integration of older migrants and their utilization of public health services. Older individuals with higher levels of education may possess a strong self-awareness that hindered their cultural integration within the community (31). On the other hand, individuals with lower levels of education may find it easier to adapt to the local culture but may lack awareness of medical facilities and national policies, leading to suboptimal utilization of public health services. The utilization of public health services by the older adult floating population is positively influenced by the other three dimensions and overall social integration. Previous studies have shown that embracing public health services can improve social integration (32), indicating a mutually reinforcing relationship between social integration and public health services. Enhancing social integration can improve the access to public health services for the older adult floating population, which in turn can enhance their sense of identity and social integration. It is crucial to leverage this connection to continuously enhance relevant support measures and improve the social integration of the older adult floating population while also enhancing public health services.

The utilization of public health services among older adult migrants is influenced by various factors, as evidenced by multivariate analysis. Notably, senior males with at least a junior high school education are more likely to belong to class 2 rather than the class 1. This suggests that higher levels of education can influence access to healthcare services for older adult male migrants. In a previous study, it was observed that women in the migrant population were more receptive to health education compared to men (33). However, the present study revealed an opposite trend. This difference could be attributed to varying health education needs between younger and older individuals. Prior research has demonstrated that women prioritize maternal and child healthcare, eugenics, and contraception in health education acceptance, while older individuals prioritize prevention and treatment of chronic diseases. It has been suggested that the reduced emphasis on maternal and child healthcare among older women may contribute to their lower acceptance of health education (34). Similarly, older adult individuals who relocate alone are more likely to belong to the higher class. This could be attributed to the fact that older individuals who move without family or friends in their new area tend to connect more with the local community over time. Consequently, they become more familiar with local public health resources, facilitating their access to public health services. This underscores the positive impact of social networks and integration within one's place of residence on the utilization of public health services. Conversely, older adult individuals who move to different municipalities or provinces often encounter challenges in accessing public health services due to increased mobility, significant changes in living and cultural environments (35), and potential language barriers (36). Their lack of familiarity with local services hinders their effective utilization of healthcare resources, making integration into the local community and accessing public health services more difficult. Overall, these findings emphasize the significance of education, social integration, and familiarity with local resources in determining the utilization of public health services among older adult migrants. Addressing these factors can help improve access and overall health outcomes for this vulnerable population.

It is important to acknowledge several limitations of the study. Firstly, the cross-sectional design used in this study limited the ability to establish causal relationships. Future research employing longitudinal designs would be valuable in exploring the stability of the identified classes over time. Secondly, relying on participants' recollection for reporting their use of public health services introduced the possibility of recall bias, potentially affecting the accuracy of the data. The use of objective measures, such as medical records or administrative data, could enhance the reliability of the findings. Furthermore, the subjective nature of the criteria used to measure social integration might introduce bias and affect the fairness and accuracy of the results. Future studies could incorporate validated measures or employ multiple indicators to provide a more comprehensive assessment of social integration. In addition, due to the limitations of the database itself, many variables that may have an impact on the utilization of public health services by the older adult migrant population were not included in this study. For example, the lifestyle of the older adult as well as their medical habits may have a direct impact on their utilization of public health services, and it is suggested that future studies may explore these variables. It is also important to recognize that the study focused specifically on the older adult migrant population in China. Therefore, it remains unclear whether these findings can be generalized to other demographic groups or populations. Future research should address these limitations and explore various subgroups within the migrant population to gain a more comprehensive understanding of social integration and public health service utilization.

5 Conclusion

The study identified three distinct classes of public health service utilization among the older adult mobile population: low, medium, and high. The high utilization class was more likely to include older migrants who were male, had higher education, moved alone, and had a relatively short distance of migration. Furthermore, the findings indicated that good social integration was associated with increased utilization of public health services, whereas cultural integration seemed to hinder access to these services. To enhance the utilization of public health services among older adult migrants, it is crucial for relevant authorities to tailor these services to meet the specific needs of this population. A key strategy would be to prioritize the promotion of social integration among older adult migrants.

Data availability statement

Publicly available datasets were analyzed in this study. This data can be found at: https://www.chinaldrk.org.cn/wjw/#/data/classify/population/yearList.

Author contributions

XB: Writing – original draft, Writing – review & editing, Conceptualization, Data curation, Software. TW: Conceptualization, Data curation, Writing – review & editing, Writing – original draft. QD: Conceptualization, Software, Writing – review & editing, Writing – original draft. CL: Conceptualization, Software, Supervision, Writing – review & editing, Writing – original draft.

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Conflict of interest

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

Publisher's note

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EDITED BY
Gustavo Cunha de Araujo,
Federal University of North Tocantins (UFNT),
Brazil

REVIEWED BY
Baiba Martinsone,
University of Latvia, Latvia
Barbara Sawicka,
University of Life Sciences of Lublin, Poland

*CORRESPONDENCE
Helen Keyes
✉ helen.keyes@aru.ac.uk

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Creating arts and crafting positively predicts subjective wellbeing

Helen Keyes*, Sarah Gradidge, Suzanna Elizabeth Forwood, Nic Gibson, Annelie Harvey, Evelin Kis, Karen Mutsatsa, Rachel Ownsworth, Shyanne Roeloffs and Magdalena Zawisza

Applied Social Change Hub, School of Psychology and Sport Science, Anglia Ruskin University, Cambridge, United Kingdom

Introduction: This study explored whether engagement with Creating Arts and Crafting (CAC) predicted subjective wellbeing and loneliness, above and beyond known sociodemographic predictors.

Methods: Secondary data from 7,182 adults living in England from the Taking Part Survey (a 2019–2020 UK household survey of culture and sport participation) were analyzed. Hierarchical Linear regressions were used to explore the predictive effect of engagement with CAC on aspects of subjective wellbeing (anxiety, happiness, life satisfaction and a sense that life is worthwhile) and loneliness. Covariates included gender, Index of Multiple Deprivation (IMD), age group, health and employment status.

Results: Engaging in CAC significantly predicted increased life satisfaction, a sense that life is worthwhile and happiness, above and beyond known sociodemographic predictors.

Conclusion: Our study provides support for the wellbeing benefits of engagement with creating arts and crafting, and we suggest that this will be a useful tool at a public health level, noting that relative accessibility and affordability creating arts and crafting.

KEYWORDS

wellbeing, arts and crafts, life satisfaction, happiness, worthwhile life

1 Introduction

Since the COVID-19 pandemic, mental health provisions are failing to meet the growing demand on their services from people in need (1). Wellbeing describes how positive an individual's psychological state is, which encompasses both feeling well [positive emotions and life satisfaction; (2)] and doing well [a sense of fulfillment; (3)]. Positive wellbeing is associated with a multitude of beneficial outcomes, including higher educational achievement (4), better physical health outcomes (5), more positive physical health behaviors (6) and reduced mortality (7). Loneliness is a negative psychological experience as a consequence of a deficit in high quality social connection (8, 9), and is associated with lower quality of life (10), as well as negative health behaviors and outcomes, such as substance misuse, smoking, eating disorders, depression, stress and premature death (8, 9). Any interventions to combat the profound public health issues of low wellbeing and high loneliness could therefore have a significant impact.

One suggested tool to improve wellbeing and combat loneliness is engagement with Creating Arts and Crafting (CAC). CAC can refer to numerous activities, such as pottery, drawing, painting, knitting, sewing, and crochet. There is wide-scale public interest in CAC, with British TV programs, such as *The Great British Sewing Bee* and *All that Glitters* populating prime time television slots. Approximately 20% of the British population are estimated to engage in arts and crafts (11). The arts and crafts industry contributes about £3.4 billion to the UK economy (12) and is forecast to be worth $50.91 billion worldwide by 2024 (13). Given its popularity, the unique purpose of this paper is to address gaps in the literature and directly assess the benefits of engagement in CAC on subjective wellbeing and loneliness, above and beyond known sociodemographic variable predictors within the general population.

Engagement with CAC is not a new intervention to improve mental health and wellbeing in clinical populations [e.g., (14)]. Indeed, CAC has been found to help with mental health issues (14, 15), enhance subjective wellbeing (16–20) and reportedly reduce suicidal tendencies (21). Therefore, not only is engagement with CAC of public interest, but it may also bring profound wellbeing benefits. As such, it has potential to be an optimal wellbeing intervention. However, while a substantial body of work has demonstrated the positive outcomes of engaging with CAC, most studies have investigated therapeutic interventions in clinical populations, only explored specific types of crafts and/or utilized small sample sizes, often with a qualitative approach [e.g., (14, 22–28)]. Thus, the extent to which the broad spectrum of CAC activities quantitatively contributes to wellbeing and loneliness outcomes in the everyday lives of the general population is comparatively understudied, and is the focus of this paper.

Less studied is whether engagement with CAC might be associated with reduced loneliness. Mindfulness is known to be effective in reducing loneliness [e.g., (29)], and there are parallels between mindfulness and CAC; motivations to engage in creative activities, including crafting, often center around their use as a tool to disengage from unwanted thoughts and feelings and to direct and focus attention onto the task at hand (30). This has been applied in therapies that use arts to facilitate mindfulness (31, 32). Considering the parallels between mindfulness and people's motivations to engage in creative tasks, we seek to investigate here whether engagement with CAC can be effective in reducing loneliness.

1.1 Research question, aims and hypothesis

The current study aims to address these gaps in the literature. To this end we utilize a dataset from the Taking Part survey, an annual survey conducted by the UK Department for Culture, Media and Sport, which assesses a sample of the general population's engagement in cultural, digital and sporting activities. We aim to quantitatively investigate whether engagement with *general* (rather than specific) CAC acts as a protective factor to enhance subjective wellbeing and reduce loneliness in a general (rather than clinical) population in a large, representative sample.

The current study controls for sociodemographic variables that are already known to be linked to subjective wellbeing and loneliness outcomes (33): level of deprivation, gender, age group, health and employment status. For instance, lower wellbeing and greater loneliness have been linked to greater deprivation (34, 35), poorer health (36, 37), and unemployment (38, 39). Loneliness also increases with age (40–42), with subjective wellbeing showing a U-shaped curve in high income countries, rising from the 45–54 age range onwards (36). Studies on gender differences in wellbeing and loneliness are more mixed [e.g., (42–45)] and sometimes yield only small effect sizes [e.g., (44)]. We include these sociodemographic variables in our analyses to: (a) control for statistical relationships between these known sociodemographic predictors and wellbeing/loneliness, and (b) to assess the relationship between engagement with CAC and wellbeing/loneliness *above and beyond* these known sociodemographic predictors. The latter enables us to determine by how much engagement with CAC contributes to subjective wellbeing and loneliness, in comparison to known sociodemographic predictors and their effect sizes. This assessment of magnitude allows us to evaluate how beneficial engagement with CAC may be to wellbeing and loneliness in a real world setting.

Wellbeing is measured here through Subjective Wellbeing (SWB), which comprises the variables of happiness, anxiety, life satisfaction and a sense that life is worthwhile, in line with the UK Office for National Statistics [ONS; e.g., (46)], as these variables provide a good measure of both feeling well [positive emotions and life satisfaction; (2)] and doing well [a sense of fulfillment; (3)]. The ONS has previously outlined the rationale for using these questions to measure subjective wellbeing (47). Our key research question asks: Does engagement with CAC predict SWB and loneliness *above and beyond* sociodemographic predictors of gender, deprivation, age group, health and employment? We predict that engagement with creating arts and crafting (vs. no engagement) will be associated with greater SWB and lower loneliness, over and above known sociodemographic predictors.

2 Methods

2.1 Participants

A random representative sample of 7,182 individuals were derived from the Taking Part Survey, a face-to-face household survey undertaken prior to COVID, in Year 13 (April 2019–March 2020), by the Department of Digital, Culture, Media and Sport (48). This dataset is available from the UK Data Service (48). The participants were aged 16 and over and living in England. Although the original dataset has 7,502 participants, the final sample size used in the main analyses for the current study was 7,182 due to missing data on outcome variables and/or covariates from 320 participants. Table 1 reports participants' characteristics.

2.2 Measures

2.2.1 Sociodemographics

As per Table 1, participants reported sociodemographic data regarding their age (categorical), gender (binary: *male* vs. *female*) and home postcode. The latter informed an index of multiple deprivation (IMD (49, 50)). IMD ranges from 1 to 10 where 1 represents the relatively most and 10 least deprived deciles, respectively. Participants

TABLE 1 Participant characteristics.

Characteristic	*N* (%) (Total *N*=7,182)
Gender, *N* (%)	
Female	3,902 (54.3%)
Male	3,280 (45.7%)
Age group, *N* (%)	
Not reported	36 (0.5%)
16–19	21 (0.3%)
20–24	172 (2.4%)
25–34	925 (12.9%)
35–44	1,229 (17.1%)
45–54	1,291 (18%)
55–64	1,285 (17.9%)
65–74	1,223 (17%)
75–84	756 (10.5%)
85+	244 (3.4%)
IMD decile (scale = 1–10), *N* (%)	
1 (relatively most deprived decile)	732 (10.2%)
2	722 (10.1%)
3	613 (8.5%)
4	650 (9.1%)
5	772 (10.7%)
6	707 (9.8%)
7	770 (10.7%)
8	775 (10.8%)
9	778 (10.8%)
10 (relatively least deprived decile)	663 (9.2%)
Self-reported poor health (scale = 1–5), mean (SE)	2.11 (0.012)
In employment	
Yes, *N* (%)	3,995 (55.6%)
No, *N* (%)	3,187 (44.4%)
Engagement with creating arts and crafting	
Yes, *N* (%)	2,689 (37.4%)
No, *N* (%)	4,493 (62.6%)
Satisfaction (scale = 1–10), mean (SE)	7.76 (0.022)
Happy (scale = 1–10), mean (SE)	7.64 (0.025)
Worthwhile (scale = 1–10), mean (SE)	8.00 (0.021)
Anxiety (scale = 1–10), mean (SE)	2.79 (0.035)
Lonely (scale = 1–5), mean (SE)	2.22 (0.014)

Index of multiple deprivation (IMD), Number (N), Standard error (SE).

were also asked to self-report their health (‘*How is your health in general?*’, 1–5, higher scores indicate poor health) and employment status (‘*Are you working?*’, options: ‘*working*’ vs. ‘*not working*’).

2.2.2 Engagement with creating arts and crafting

To capture this concept, we compared participants who indicated they engaged in at least one of the following activities within the last 12 months (vs. none): ‘*Painting, drawing, printmaking or sculpture*’ (question artp13), ‘*Photography as an artistic activity (no family or holiday ‘snaps’)*’ (artp14), ‘*Made films or videos as an artistic activity (not family or holidays)*’ (artp15), ‘*Used a computer to create original artworks or animation*’ (artp16), ‘*Textile crafts such as embroidery, crocheting or knitting*’ (artp17), ‘*Wood crafts such as wood turning, carving or furniture making*’ (artp18) and ‘*Other crafts such as calligraphy, pottery or jewellery for yourself*’ (artp19). Specifically, we used the variable artp263Y13 (whereby participants were asked to indicate if they participated in ‘none of these’ activities) and reversed coded this variable for the current study, so that one indicated a participant had taken part in at least one craft activity, and zero indicated a participant had not taken part in any of these craft activities. Overall, 37.4% of participants in the survey had engaged in at least one craft activity within the past 12 months, while 62.6% had not.

2.2.3 Subjective wellbeing (SWB)

Subjective wellbeing was measured through four single-item questions, whereby each was assessed on 0–10 Likert scales and analysed individually following ONS guidance (44). These items were: life satisfaction: “*Overall, how satisfied are you with your life nowadays*” (0 = “*not at all satisfied?*” to 10 = “*completely satisfied*”), life being worthwhile: “*To what extent do you feel that the things in your life are worthwhile?*” (0 = “*not at all worthwhile*” to 10 = “*completely*”), happiness: “*Taking all things together, how happy would you say you are?*” (0 = “*extremely unhappy*” to 10 = “*extremely happy*”), and anxiety: “*On a scale where 0 is “not at all anxious” and 10 is “completely anxious,” overall, how anxious did you feel yesterday?*” (0 = “not at all anxious” to 10 = “completely anxious”).

2.2.4 Loneliness

A single item measured loneliness: “*How often do you feel lonely?*” where 1 = ‘*often or always*’, 2 = ‘*some of the time*’, 3 = ‘*occasionally*’, 4 = ‘*hardly ever*’ and 5 = ‘*never*’. This variable was reverse-coded for the current study, so that higher scores reflect greater loneliness.

3 Results

3.1 Analyses

To test our hypothesis, we ran hierarchical linear regressions, with engagement with CAC and sociodemographics (gender, level of deprivation, age, general health and employment status) as the predictor variables, and SWB (life satisfaction, life being worthwhile, happiness and anxiety) and loneliness as the outcome variables. Therefore, a total of five hierarchical regressions were conducted, one on each outcome variable. Sociodemographic variables were entered in Block 1, with engagement with CAC entered in Block 2. All analyses were conducted via Jamovi. The categorical predictor variables were dummy coded: engagement with CAC (reference: none of these), gender (reference: female), and employment status (reference: not working). There was no multicollinearity between the predictor variables, as assessed through VIFs ≤1.37.

3.1.1 Life satisfaction

Engagement with creating arts and crafting (CAC) significantly predicted participants’ reported life satisfaction, above and beyond the effects of age, gender, deprivation, poor health, and being in work. Inclusion of engagement with CAC in the model explained an additional 0.1% of variance in life satisfaction scores compared to the effects of a model including only age, gender, deprivation, poor health and being in work, F (1, 7,175) = 4.59, p = 0.032. Engagement with CAC predicted a greater increase in life satisfaction (β = 0.088) than the increase associated with living in a less deprived area (β = 0.018). In the final stage of the model, deprivation, age, poor health and engaging with CAC were all significant predictors of life satisfaction, with those in less deprived areas, older age groups, those in better health and those who had engaged in CAC in the last year all reporting higher life satisfaction scores. The final model accounted for 16.4% of the variance in life satisfaction scores, R^2 = 0.164, F (6, 7,175) = 235, p < 0.001. These findings align with our hypothesis.

3.1.2 Sense of life being worthwhile

Engagement with CAC significantly predicted participants' sense that life is worthwhile, above and beyond the effects of age, gender, deprivation, poor health and being in work. Indeed, inclusion of engagement in CAC in the model explains an additional 0.4% variance in participants' sense that life is worthwhile compared to a model that only includes age, gender, deprivation, poor health and being in work, F (1, 7,175) = 27.5, p < 0.001. Participants' engagement with CAC in the last 12 months had a larger effect on their sense that life is worthwhile (β = 0.218) than being in employment (β = 0.136), than aging by one decile (~ 20 years; β = 0.082) or than living in a less deprived area (β = 0.019). In the final model, all six variables significantly predicted participants' sense that life is worthwhile, with women, those in older age groups, those living in less derived areas, those reporting better health, those in employment and those who had engaged with CAC in the last 12 months all reporting a greater sense that life is worthwhile. The final model accounted for 11.2% of variance in scores measuring a sense that life is worthwhile, R^2 = 0.112, F (6, 7,175) = 150, p < 0.001. This is in line with our hypothesis.

3.1.3 Happiness

Participants' reported happiness was significantly predicted by their engagement with CAC, above and beyond the effects of age, gender, deprivation, poor health and being in work. An additional 0.1% of variance in participants' happiness scores could be explained by the inclusion of engagement with CAC in the model, compared to the effects of age, gender, deprivation, poor health or being in work, F (1, 7,175) = 6.71, p = 0.010. Engagement with CAC predicted a similar increase in happiness (β = 0.128) as aging by one decile (~ 20 years; β = 0.127). In the final stage of the model, gender, age, poor health and engagement with CAC all significantly predicted happiness scores, with women, those from an older age group, those reporting better health and those who had engaged with CAC in the last 12 months all reporting being happier. The final model accounted for 10.6% of variance in participants' happiness scores, R^2 = 0.106, F (6, 7,175) = 142, p < 0.001. These findings align with our hypothesis.

3.1.4 Anxiety

Contrary to our hypothesis, engagement with CAC did not provide additional predictive power for anxiety scores to the model (p = 0.108). The final model accounts for 7.1% variance in anxiety scores, with women, those in younger age groups, and those in poorer health reporting higher anxiety scores, R^2 = 0.071, F (6, 7,175) = 91.5, p < 0.001.

3.1.5 Loneliness

Contrary to our hypothesis, a model including engagement with CAC did not predict significantly more variance in loneliness scores than a model including age, gender, deprivation, poor health and being in work alone (p = 0.805). The final model accounted for 7.7% of variance in loneliness scores, with men, older age groups, those living in less deprived areas, those in better health and those in employment all reporting being less lonely, R^2 = 0.077, F (6, 7,175) = 99, p < 0.001.

3.2 Summary

Our findings support aspects of our hypothesis; specifically that engagement with CAC is associated with significantly higher life satisfaction, happiness, and a sense that life is worthwhile. Engagement with CAC is not associated with lower levels of anxiety or loneliness (Table 2).

4 Discussion

Given the concern for improving the population's wellbeing (1) and decreasing loneliness (51), this paper set out to uniquely and directly assess the benefits of engagement with CAC to combat poor wellbeing and loneliness. For the first time, our paper demonstrates that engaging in general (rather than specific) crafting and arts activities may be beneficial in improving subjective wellbeing, over and above one's gender, health, age, employment status and affluence. Importantly, we show that this pattern holds for the general (rather than clinical) population using quantitative (rather than qualitative) methods, addressing several gaps in the literature.

Specifically, in line with our hypothesis, engagement with CAC was linked to significantly increased life satisfaction, a greater sense that life is worthwhile and increased happiness, above and beyond known sociodemographic predictors. While these findings are of a small-sized magnitude, they are comparable to the effects of known sociodemographic variables, and are of practical importance at a population level. For example, the predictive effect of engagement with CAC on life satisfaction that we observed is an order of magnitude greater than the effect of living in a less deprived area. Similarly, the predictive effect of engagement with CAC on people's sense that life is worthwhile is greater than the individual effect of being in employment, greater than the effect of living in a less deprived area, and greater than the effect of aging by one decile (~ 20 years). The effect of engagement with CAC on happiness was of a similar magnitude as the effect of aging by one decile or of being female. That is, although variances explained by engaging with CAC were objectively low, they are relatively larger (for life satisfaction and life being worthwhile) or comparable (for happiness) than those explained by other sociodemographic variables known to impact wellbeing. The relationships between engagement with CAC and subjective wellbeing measures in relation to sociodemographic predictors is noteworthy, because unlike static sociodemographic variables, engagement with CAC is a variable that can be manipulated and is therefore open to

TABLE 2 **Results of two-step hierarchical multiple regressions predicting satisfaction, happiness, life being worthwhile, anxiety and loneliness with regression coefficients (B) specified for all predictor variables at each step of the regression.**

Predictor variables	Satisfaction	Happiness	Worthwhile	Anxiety	Loneliness
Step 1					
Intercept	8.694***	8.418***	8.682***	2.559***	2.183***
Gender	−0.071	−0.145**	−0.234***	−0.387***	−0.209***
IMD decile	0.019**	0.012	0.021**	0.021	−0.019***
Age group	0.100***	0.126***	0.082***	−0.239***	−0.048***
General health	−0.749***	−0.697***	−0.571***	0.753***	0.284***
In employment	0.062	0.019	0.136**	0.002	−0.187***
R^2	0.163	0.105	0.108	0.071	0.077
F	280***	169***	174***	109.3***	118.8***
Step 2					
Intercept	8.653***	8.358***	8.580***	2.506***	2.187***
Gender	−0.063	−0.128**	−0.205***	−0.371***	−0.210***
IMD decile	0.018*	0.011	0.019**	0.020	−0.019***
Age group	0.100***	0.127***	0.083***	−0.238***	−0.048***
General health	−0.747***	−0.695***	−0.567***	0.755***	0.284***
In employment	0.062	0.018	0.136**	0.002	−0.187***
ECAC	0.088*	0.128*	0.218***	0.113	−0.007
R^2	0.164	0.106	0.112	0.071	0.077
ΔR^2	0.0005	0.0008	0.003	0.0003	<0.0001
ΔF	4.59*	6.71*	27.5***	2.59	0.061

*$p < 0.05$; **$p < 0.01$, ***$p < 0.001$.

intervention. Thus, not only is engagement with CAC possibly more effective than many sociodemographic variables on improving aspects of wellbeing, but it is also an easier variable to manipulate and influence. We note also that engagement with crafting or creating arts has a relatively low entry point; crafting and creating arts is an accessible and relatively affordable pastime, and a popular pursuit among the general population. This is in contrast to attending live sporting events, which has also been demonstrated to positively predict SWM in a large UK sample (33), though attending live sporting events predicted lower loneliness, whereas engagement with CAC did not.

The implications of these findings are particularly significant given the importance of wellbeing. Higher SWB predicts health (52) and longevity (53), with some evidence pointing to positive effects on those with certain illnesses (54). Diener and Chan (55) outline the causal evidence demonstrating the beneficial influence SWB has on health and longevity. They also suggest policy makers include interventions to improve SWB across the population, given the potential impact of small SWB increases. Engagement with CAC is related to three of the four SWB measures assessed in this study. So, while the effects may be small, the contribution to SWB as measured by life satisfaction, happiness and life being worthwhile may provide a meaningful influence across society as a whole.

Our hypothesis was not supported with regards to one component of SWB (anxiety) and loneliness, as engagement with CAC had no significant predictive effect on either, above and beyond other sociodemographic predictors. Arts and crafts activities included here (e.g., drawing, painting, knitting, sewing, and crochet) can be considered as relatively solitary activities. This likely explains why we did not observe an effect of engaging with CAC on reducing loneliness. Further research should unpick the importance of the social elements of engagement with CAC, especially given the rich literature on social support and wellbeing (56).

Regarding the role of sociodemographic factors on SWB and loneliness, we largely replicate findings from previous research, as expected. We found that loneliness and perceiving life as worthwhile were both predicted by all sociodemographic variables here (deprivation, age group, gender, general health and employment status). Life satisfaction was predicted by deprivation, age and general health, whereas happiness and anxiety were both predicted by gender, age and health. In general, better health (36, 37) and older age (36, 40–42) are predictive of decreased loneliness and higher SWB. Further, higher levels of life satisfaction and the sense that life is worthwhile were associated with living in less deprived areas, as shown in previous research (34, 35). Similarly, loneliness is reported as lower in less deprived areas (57, 58). Previous literature also supports employment status being linked to factors associated with enhanced SWB (38, 59) and decreased loneliness (39). We found gender was not associated with life satisfaction, and previous literature has also been mixed in this regard (42–44, 57).

While our study's strengths include the use of a nationally representative sample, investigation of overall engagement in craft activities and the use of quantitative methods, it is not without limitations. Firstly, our list of arts and crafts is not exhaustive and other arts categories, such as metalworking and literary arts were not considered. We also cannot be sure that participants shared the same understanding of what the various crafts listed in the survey entail. Of interest, many of the crafts included here may be seen as stereotypically feminine and thus the choice of CAC activities may be confounded with gender as a sociodemographic. It is unclear whether different arts and crafts affect men and women differently. Literature on masculinity threat and precarious manhood suggests that crafts such as knitting may be seen as threatening for many men (60). This question awaits further investigation, in particular whether stereotypically 'masculine' crafts (e.g., carpentry, metalworking) may have a larger impact on men's wellbeing (29, 30). Secondly, it is important to note that the present study was correlational and therefore while we can speculate on the impact of engaging with CAC on life satisfaction, causation cannot be determined. The next step for future research is to experimentally manipulate whether or not participants engage in CAC and for how long to measure the causal impact on life satisfaction.

Further studies should also focus on the mechanisms through which engagement with CAC relates to wellbeing, which were not measured here. For example, a key component of creativity is the experience of flow (61), an intrinsically rewarding state characterized by total immersion in an activity. Frequent flow experiences have been linked to better quality of life, in particular wellbeing, satisfaction and a sense of mastery (62, 63). Creative arts are also associated with improved self-esteem and emotion regulation, and provide a means of authentic self-expression (64–70). A range of studies show that engagement in art therapy results in a reduction in anxiety (71), depression (72) and stress (73, 74), and improvements in social connection, wellbeing and life satisfaction (27, 68, 75, 76). Determining the exact mechanisms through which engagement with CAC may be beneficial for well-being awaits further studies.

Overall, our study provides support for exploring the wellbeing benefits of engagement with creative arts and crafting as a useful strategy to improve wellbeing at a population level, with the positive effects observed here being comparable to or greater than known sociodemographic predictors of wellbeing. Being already popular (37.4% of the current national sample), relatively cheap and accessible, engagement with CAC activities lends itself to government support and public uptake. Increased funding for creative arts and crafting activities for the general population may benefit society as a whole by improving wellbeing in modern living.

Data availability statement

Publicly available datasets were analyzed in this study. This data can be found here: https://beta.ukdataservice.ac.uk/datacatalogue/doi/?id=8745#!#0.

Ethics statement

Ethical approval was not required for the study involving humans in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and the institutional requirements.

Author contributions

HK: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. SG: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. SF: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft. NG: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft. AH: Writing – review & editing. EK: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. KM: Writing – review & editing. RO: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. SR: Conceptualization, Investigation, Writing – original draft. MZ: Conceptualization, Formal analysis, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing.

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Olatunde Aremu,
Birmingham City University, United Kingdom

REVIEWED BY
Adewale Adebajo,
The University of Sheffield, United Kingdom
Edford Sinkala,
University of Zambia, Zambia

*CORRESPONDENCE
Fiona Borondy-Jenkins
✉ Fiona.bjenkins@hepb.org

†These authors have contributed equally to this work

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Global hepatitis B and D community advisory board: expectations, challenges, and lessons learned

Fiona Borondy-Jenkins*†, Bright Ansah, Jacki Chen, Amanda Goldring, Yasmin Ibrahim†, Shaibu Issa, Silvana Lesidrenska, Tanya Machado†, Holly Moore†, Richard Njouom, Prince Okinedo, Rhea Racho, Lori Scott, Beatrice Zovich† and Chari Cohen†

Hepatitis B Foundation, Doylestown, PA, United States

Introduction: Community Advisory Boards (CABs) play an important role in developing and delivering patient-centered care. However, the impact of participation on CAB members has not been well studied, particularly on the global scale. In 2022, the Hepatitis B Foundation (HBF) convened the first global hepatitis B and hepatitis delta CAB with 23 members from 17 countries, representing six out of the seven World Health Organization (WHO) regions, and countries with the largest hepatitis B and hepatitis delta disease burden.

Methods: To reflect on the process of assembling an effective and motivated CAB and assess the impact on CAB participants, three virtual focus group sessions were held with 16 participants in July and August 2023. Sessions were recorded and transcribed. Questions focused on motivations for joining the CAB, membership experiences, and lessons learned. Grounded theory analysis was used to generate hypotheses about reasons for CAB members' participation, as well as challenges and suggestions. Qualitative analysis using inductive reasoning identified key themes within responses. Transcripts were independently analyzed by a primary and secondary coder.

Results: Motivations for joining the CAB included participants' desire to advocate for people living with hepatitis B and hepatitis delta, and other altruistic factors. Participants reflected that through CAB membership, they gained networking and advocacy opportunities and enhanced their hepatitis B- and hepatitis delta-related knowledge. Challenges participants experienced were related to time, physical limitations, and stigma. Finally, participants discussed their limited direct engagement with drug developers and proposed ways the CAB can increase interactions with stakeholders going forward.

Discussion: Based on participants' assessments, establishing a global CAB for stigmatized infectious diseases is worth the effort. Regular internal review of community advisory boards' structure and performance is critical to ensure the CAB is fulfilling its mission.

KEYWORDS

community empowerment, research participation, global advisory board, clinical trial design, drug development, hepatitis B, hepatitis D, patient centricity

1 Introduction

According to the World Health Organization (WHO), there are 254 million people living with chronic hepatitis B virus (HBV) (1), and an estimated 12.7 million people living with hepatitis delta (HDV) worldwide (2). Hepatitis B is the most common serious liver infection in the world, but is preventable with a vaccine, and treatable with drug therapies. HBV heavily affects those in high endemic areas including in some parts of Asia, the Western Pacific, Africa and Eastern Mediterranean countries (1, 3). Hepatitis delta affects between 5 and 10% of individuals living with HBV. Because HDV depends on HBV to survive and reproduce, it is also preventable with the hepatitis B vaccine. Although HDV is less common, it is the most severe form of viral hepatitis and is seen most frequently in Mongolia, Moldova, Romania, and countries in Western and Central Africa (4). In the United States, both HBV and HDV are more frequently seen in populations that were born in regions of high endemicity (5).

Historically, patient preferences and insight have not been rigorously included in the development of HBV/HDV clinical trials. However, with many new HBV/HDV clinical trials being initiated, now is the ideal time to engage with people impacted by HBV/HDV. In keeping with researchers' efforts to design precise and holistic clinical trials, patient experiences must be incorporated to ensure outcomes are appropriate and effective (6). This can only be achieved by engaging people with lived experience of the disease in the entire process, from conceptualization and design to follow-up. Collaborative efforts between researchers and affected populations are necessary to identify and address key clinical needs that are most relevant to patients.

Community advisory boards (CABs) provide a well-established structure for community engagement through fostering collaborative partnerships with researchers to integrate patient perspectives on pertinent issues (7). Patient insights are crucial for enhancing the design and acceptance of clinical trials (8) and ensuring potential participants' comprehension of research practices, such as the informed consent process (7). CABs offer community members a platform to express concerns and recommend research approaches that are respectful to their community (9). CABs tailored for diseases such as HIV/AIDS (10, 11) and tuberculosis (12) have demonstrated success in promoting participatory research and designing person-centered clinical trials that align with the preferences of affected communities. Limited documented strides have been made in the areas of HBV and HDV.

In 2022, the Hepatitis B Foundation (HBF) established a global CAB, composed of people living with hepatitis B and delta (PLHB and D), as well as caregivers and advocates, to increase engagement between these communities and those working in the drug development and clinical trial spaces, and to amplify the voices of those most affected by these viruses. Upon conception of the CAB, the research team sent out an open call for applications that was widely distributed through social media channels, email listservs and website postings, as well as through an established network of patients, providers and community advocates in the HBV/HDV space. Those interested were invited to apply to join the CAB. Founding members were selected based on certain qualifications, including but not limited to their HBV/HDV status or being a caregiver to someone living with HBV/HDV, English proficiency and their experience in HBV/HDV education and advocacy efforts. Joining the CAB is voluntary, and those who chose not to submit an application were not considered to join. As the founding cohort of CAB members are nearing the end of their first two-year term, HBF sought to document the process of assembling a highly involved CAB, including members' motivations for joining, their membership experiences, and any lessons learned regarding their interactions with drug developers and other stakeholders.

In contrast to most existing literature about the role of CABs, which focuses on the healthcare space, this qualitative study presents the benefits and challenges of a global CAB within the public health and clinical research space, as well as potential suggestions for deepening engagement. Documenting the inaugural cohort's experiences and the initial challenges, can provide guidance the current CAB's sustainability efforts and to others seeking to embark on similar endeavors to build robust cohorts of global advocates who can transform the drug development process into one that is truly patient-centered.

2 Methods and materials

In July and August 2023, three virtual, 90-min focus group sessions were facilitated by trained moderators in English, using Zoom. All CAB members were invited to participate. Of those who participated in the focus groups, there were 10 HBV and six HDV CAB members. Two experienced, external third-party moderators facilitated the three focus groups to reduce the risk of social desirability bias. One researcher joined each focus group as a silent observer, to provide technical support and take notes during each session. With participant consent, sessions were audio-recorded and professionally transcribed.

Grounded theory analysis was used to generate hypotheses about participants' anticipated responses. This is a qualitative research method in which empirical data is collected first, and then researchers create a theory 'grounded' in the results of the data. This is often used to analyze social processes (13). This method informed the creation of a focus group discussion guide (Supplementary Figure 1) (13). There were five sections in this guide, spanning their understanding about the CAB's goals, their motivations for joining the CAB, their experiences, their thoughts about drug development and any lessons learned.

Qualitative analysis using inductive reasoning was used to identify key themes within responses, which along with the guide, informed the creation of a codebook (Supplementary Table 2). Each transcript was independently coded by a primary and secondary coder using the qualitative analysis software NVivo Version 13 (14). Intercoder agreement, or reliability, is measured by computing for the Kappa coefficient, a statistical test that offers the measure of agreement between different coders' categorization of data and controls for the random agreement factor (15, 16). The final kappa coefficient was 0.80, indicating high inter-coder reliability.

2.1 Ethical considerations

Prior to their participation, participants were made aware of the possibility that their identity might not remain confidential, as their membership in the CAB is public information and thus disclosure of HBV/HDV status was a potential risk of focus group participation. This study was approved by Heartland Institutional Review Board (HIRB) (HIRB Project No. 081122-407).

3 Results

3.1 Participant characteristics

Across the three focus groups, there were 16 participants who represented 11 different countries. Table 1 presents the demographic characteristics of participants.

3.2 Findings

Additional quotes for each theme can be found in Supplementary Table 1.

3.2.1 Motivations for joining the CAB

Participants expressed many motivations for joining the CAB; representing PLHB and D, altruism [participants' motivation to do something to benefit others, rather than, only themselves (17)], and educating their community were most frequently cited as the primary motivators.

3.2.1.1 Representing PLHB and D

Most participants agreed that they wanted to be the voice of PLHB and D, to bring patients' perspectives to the forefront in drug development, policy, and advocacy efforts. Many also expected to raise awareness of the challenges these groups face worldwide. In the words of one participant from the U.K.:

TABLE 1 **Participant demographics, 2023.**

Demographics	Totals *N* (%)
Age range	
25–35	4 (25%)
36–45	5 (31.25%)
46–55	3 (18.75%)
56–65	2 (12.5%)
65 and above	2 (12.5%)
Gender	
Male	11 (68.75%)
Female	5 (31.25%)
Ethnicity	
Asian	4 (25%)
African	7 (43.75%)
Caucasian	3 (18.75%)
Eastern European	2 (12.5%)
Hepatitis diagnosis	
Hepatitis B	10 (62.5%)
Hepatitis delta	6 (37.5%)
Residing WHO region (country)	
African region	5 (31.25%)
Cameroon	1
Ghana	1
Nigeria	1
Tanzania	1
Uganda	1
Western Pacific region	2 (18.75%)
Australia	1
Philippines	1
European region	3 (18.75%)
Bulgaria	1
Romania	1
United Kingdom	1
Region of the Americas	6 (37.50%)
USA	6

Table represents the demographic makeup of study participants, including ethnicity and country of residence.

> *I think being a member of [the] CAB gives us that voice to speak out, not just for ourselves, but to speak out for so many people who have no voice at all… being their voice to the pharmaceutical companies and shouting loud and clear not just for ourselves but for everybody with hepatitis B [and D].*

3.2.1.2 Altruism

Altruism was another motivating factor for CAB membership. Many participants explained how they would be pleased if their advocacy efforts in the drug development process would lead to a cure for HBV and effective treatments for HDV for future generations, even if they did not personally benefit. A Romanian participant summarized this:

> *Of course, you hope to do it for yourself, for your friends, for your family. But then also for the next generations because, yeah, I wouldn't mind, if I would go away and just disappear, but then maybe the next generation or the two generations, hepatitis B and D would just be like some flus and that would be good … even if it didn't help my generation.*

Participants hoped that they would be able to improve peoples' lives through community education, to reduce stigma associated with hepatitis B and D, and to increase screening and vaccination rates. This idea was summarized by a U.S. participant (#1), *"I want to educate my family, my friends, the community about viral hepatitis, which you know, if people are educated, there'd be a lot less of it because at least for hepatitis B, there are vaccinations."*

3.2.1.3 Knowledge and skill-building

Participants also joined the CAB because they wanted to build their knowledge and improve their advocacy skills to make a difference in a global context. Specifically, participants emphasized their desire to advocate for PLHB and D by bringing more medical and research resources to the African context. One participant from Cameroon stated, *"Do not forget Africa"* when working toward hepatitis elimination efforts. A participant from Tanzania also shared, *"Who can break the silence is us. We need to tell our government that there is a problem, and we need to end this problem. And the best people with knowledge … can come from this CAB."*

3.2.2 Membership reflections

When reflecting on their membership in the CAB, participants cited positive outcomes, including building greater confidence and developing global connections, however, some have cited personal challenges.

3.2.2.1 Experience as a CAB member

Many participants highlighted increased confidence when advocating for PLHB and D, as expressed by a participant from the U.K., *"I was diagnosed with HBV three and half years ago and for a long time I felt I had no voice whatsoever. And I think being a member of [the] CAB gives us that voice to speak out."* A participant from Nigeria added:

> *Frankly, this [CAB] has really impacted positively in the way I carry on with hepatitis advocacy. I used to be this very shy person when it comes to reaching out to people. But somehow this has emboldened me and given me a lot of confidence.*

Other positive experiences included feeling empowered to raise questions and propose solutions during internal CAB discussions. For example, a participant from the Philippines said, *"The discussions are really more important to me because it is where we can raise our questions and give our suggestions and opinions."*

Many also felt that their affiliation with a global advisory board lent greater credibility to their local advocacy endeavors. One Nigerian participant articulated, *"When I do my advocacy work and I talk to people in government and I let them know that I'm a member of CAB, they really want to listen. And I think that's a huge support for me."*

Conversely, others felt the CAB is not acknowledged by pharmaceutical companies, policymakers, and government agencies even with the Foundation's support. Many felt this inhibits their ability to provide important insights about PLHB and D's preferences in drug development. A U.S. participant (#2) remarked:

> *I think the barrier is enormous. [It is not easy for us] to interact with the influential stakeholder[s] like the government agency, the policymaker. We rely on the Hep B Foundation to be able to give us a sort of a seat there that we can communicate with them. I'm hoping that some of the stakeholders will be able to pay more attention to individual patients' voices, or as a CAB member that we have more chance[s] to interact with them.*

3.2.2.2 Global team membership

Participants discussed how the CAB has equipped them with newfound global collaboration opportunities, and how this has benefitted their personal advocacy endeavors. A Nigerian participant underscored the breadth of the international relationships fostered through the CAB: *"I'll be organizing a summit in my State, and some of the CAB members will be featured as guest speakers."*

Additionally, a participant from Ghana explained how the cooperative nature of the CAB has provided them with global learning opportunities: *"I'm feeling comfortable honestly because [I am] learning from people [who are] globally bringing their experiences [and] sharing what they think."*

3.2.2.3 Building knowledge

Participants appreciated the knowledge gained through training, discussions, and presentations from guest speakers. A U.S. participant (#3) explained how the required trainings enhanced their understanding of drug development processes:

> *To me the most important aspect was having training[s] on the drug discovery process. Many of us patients, we are always concerned about when [a] treatment or cure will be available. But what we tend to forget is finding treatment[s] or finding a cure is not a straightforward line.*

3.2.2.4 Personal challenges

Some participants cited difficulties that accompany the coordination of a global group of individuals with lived experience, including identifying meeting times that are convenient for all time zones and making travel accommodations for PLHB and D, especially those who are limited due to physical ailments. One U.S. participant (#3) explained, *"I battle severe chronic fatigue, so it's hard sometimes to travel."* Additionally, there is the added level of fear about stigma and discrimination that comes with disclosing one's disease experience, as expressed by a U.S. participant (#4):

> *Why is it so difficult to be an advocate for this disease? It's because a big part of [it is] the unseen, the stigma, prejudice and discrimination. So, for me it's trying to find that yes, I want to contribute to this cause and let me share as much as I can to support that. At the same time, if this information gets shared somewhere, sometimes people are not understanding and there are consequences or risks that I end up taking.*

3.2.3 Challenges and suggestions for increasing CAB engagement with industry

Participants reflected on the barriers they faced when trying to establish relationships with industry stakeholders. Participants suggested ways the CAB could increase interaction with drug and clinical trial developers and highlighted the body's potential to contribute to the development of patient-centric clinical trials and effective drug outcomes.

3.2.3.1 Engagement with drug and clinical trial developers

Engagement with drug developers proved to be one major and unanticipated difficulty in the inaugural year of the CAB. Many participants felt that drug developers did not take them seriously, as one participant from Romania stated, *"But then when we go and interact with the [drug developers], we have to somehow be [an equal player] for them also to be taken seriously."* Another participant from Bulgaria was also eager to establish relationships with developers, *"So, if there is a possibility or some hidden keys, how to communicate with stakeholders openly and without any restriction, I'm open to hear."*

Many participants proposed ideas for cultivating stronger relationships with researchers and drug developers going forward. One recurring thought was to build a larger presence, as representatives of PLHB and D, at the conferences and events attended by drug and clinical trial developers. A Romanian participant proposed:

> *For us, what to do is just to be a powerful organization with a lot of members and be vocal and present at whatever world events we might hear about, it doesn't matter if it's [a conference] in Tanzania or in Paris, just be there with a member to have our voices and our organized ideas heard.*

3.2.3.2 Unique contributions of CAB members

Participants presented various suggestions about how their insights could be useful to those working in the drug development and clinical trial spaces. Chief among these was the simplification of language to ensure that study materials are not burdensome for clinical trial participants to understand. One U.S. participant (#3) noted, *"…the way they are written out sounds like it's written just for the pharmaceutical company and not for the patient. Because half of the thing you read is like you cannot make any sense of it."*

Additionally, participants referenced their ability to serve as ambassadors to the larger patient community, bridging the gap between scientists and PLHB and D. In particular, many noted their potential to enhance recruitment efforts by assuaging the community's apprehension of research. A Bulgarian participant remarked:

> *Many patients are afraid to participate in clinical trials… So, I think one thing we can do [is] develop materials that are in [an] understandable language just to spread the word…patients don't realize that to have [an] approved drug, a clinical trial should be done with people. I mean, they are all afraid, but they are awaiting the drug, but we need to participate in clinical trials, otherwise we don't have the drug.*

Participants felt that their dignity and personhood could be lost when taking part in a study. To overcome this, one participant from the U.K. explained how their interactions with clinical trial developers could serve as a reminder of study participants' humanity, *"I'm sure that when they are developing clinical trials, they do not necessarily see a human being at the end, if that makes sense? And [we can] represent, you know, it's a human being, it's not just a patient."*

Participants also felt that as PLHB or D and CAB members, they are in a unique position to help drug and clinical trial developers understand patient perspectives globally and create more patient-focused clinical trials. For example, when reflecting on the consequences of siloed drug development processes, one participant from Ghana posited, *"It could be one [drug] that is going to be effective in Europe [but] when you bring it in Africa it's going to work otherwise. So, I think they should also consider if that one is going to work."* Another example, discussed by a U.S. participant (#3), was helping drug developers have greater involvement in addressing the multi-faceted barriers to participation to enhance clinical trial accessibility:

> *Sometimes patients have concern with side effects, sometimes they have problems with transportation…So just having a conversation with the pharmaceutical company about all these challenges and finding a better solution that can fit in very well, I think will go a long way by having a lot of patients participate in the clinical trials.*

4 Discussion

Participants have shown enthusiasm for and dedication to being a part of the hepatitis B and delta CAB and have expressed that CAB participation has positively impacted their lives. This invokes a charge to continue working with CAB members to advance patient-centered hepatitis B and delta clinical trials and to enhance member capacity, engagement with stakeholders, and advocacy opportunities across the hepatitis B and D space.

4.1 Expectations

Collectively, CAB members shared that most expectations they had prior to joining had materialized, especially those related to internal and external opportunities for global collaboration. Many participants expected that they would be able to improve peoples' lives through community education and reduce the stigma associated with hepatitis B and D, an experience which is personal for many members. To this end, the CAB has already undertaken projects and activities to increase community awareness through education and encouraging screening, vaccination, and treatment, strategies that have been found to effectively reduce HBV transmission and mortality rates (18).

CAB members were motivated to join because of their desire to advocate for PLHB and D and to amplify patient voices within the drug development and policy spheres. Additionally, participants were interested in raising global awareness regarding the country-specific challenges faced by PLHB and D. CAB members are well equipped to do this, as they have strong support from and are trusted by their communities and have been recognized at regional and national levels for their efforts. Success in amplifying community members' voices has been documented previously, such as when an American Indian CAB raised researchers' awareness about the community's environmental challenges to healthy lifestyles prior to an intervention's implementation (19).

The CAB members, especially from countries with limited resources, are aware of the power inequity. Instead of focusing on the disparity, they have chosen to use the CAB as a platform, to leverage their association with a western organization. This alliance allows for greater support, better resources, adding value to their advocacy efforts and what they can do for their communities.

Participants emphasized their desire to contribute to finding a cure for HBV and effective treatments for HDV as motivation for joining the CAB. Many believed their advocacy efforts would help lead to a cure and treatments for future generations and were passionate in their efforts, even if these outcomes did not materialize during their lifetimes. Altruism has previously been identified as an influential factor for research participation (20, 21), and within other advocacy groups championing hepatitis C, tuberculosis and HIV treatments (22). CAB participants' dedication to helping PLHB and D illustrates how, despite being a diverse and global body, those with lived experience of a disease often share common goals which allows for membership cohesion.

4.2 Experiences

Focus group findings indicate a strong desire for those with lived experience to continue to learn, advocate, and connect through groups such as CABs. The global nature of the CAB contributed to CAB members' experiences. By leveraging the CAB as a global forum, participants can collaborate on initiatives spanning multiple countries, and nurture global networks in their educational and advocacy pursuits concerning hepatitis B and D. This collaborative atmosphere fosters a sense of community and solidarity among members, enabling them to learn from each other's diverse country-specific experiences, challenges and perspectives while navigating cultural nuances in communication and advocacy efforts across different countries. The importance of regional and global networks and replicating evidence-based models for patient advocacy has been demonstrated in other disease areas including breast cancer (23) and Pompe disease (24).

Given the goal of ultimately enhancing interactions between drug developers and patients represented by the CAB, one common theme was the value of learning the process behind treatment development and testing. Participants explained how they want to continue learning about the HBV/HDV research space, not only to improve their own advocacy skills, but also to educate their communities about the diseases. As identified in this study and previously, the capacity for individual advocacy efforts depends on accurate knowledge (25). Before joining this CAB, not all members had access to reliable information nor the chance to discuss HBV/HDV-related queries with trained healthcare professionals. One of the strengths of this CAB lies in its dismantling of this inequity by providing all members with the same learning opportunities, which has resulted in greater confidence among members when advocating for PLHB and D concerns across local and international platforms. This and previous research have illustrated the numerous ways in which patients can be involved in all aspects of the research process, and with training, can provide great support to any research initiative (26). Given participants' dedication to ongoing learning and advocacy skill-building, they are well-positioned to enhance community understanding of HBV and HDV and acceptance of possible new treatments, and possibly garner future political support for new patient-centric guidelines for screening, treatment, drug development, and care as has happened previously in the case of HIV/AIDS (27). Stronger political support from various stakeholders and increased funding availability could further encourage the insights and experiences of PLHB and D to be incorporated into further research, drug development, and access to care across the globe. Furthermore, most CAB members who participated in this study lead local and national organizations that are well recognized in their respective countries. Their advocacy and expertise will certainly contribute toward the sustainability of this CAB.

A disappointing reality of the CAB's journey so far has been difficulty engaging directly with drug developers. This has been interpreted by many members as a reluctance on the part of drug developers to consider the perspectives of those with lived experience in the research, discovery, and trial process. This experience differs from those described in existing literature, but this seems to be because much of the previous work done with CABs has included the establishment of a CAB in direct association with an existing research institute, rather than with the support of an independent disease advocacy organization, as is the case in the present study. Previously, many CABs have been established only for a particular project (28), a largely transactional experience which fails to nurture relationships between members and researchers and to center the patient voice in the drug development process. This underscores the benefits of a long-term, self-sustaining advocate group. Part of the future CAB journey will include establishing the role of this independent CAB among developers in the drug development and clinical trial spaces. Results of this assessment suggest that CAB members are already identifying possible strategies for improved future engagement, including helping researchers understand the unique benefits of engaging with an informed group of people with lived experience.

4.3 Solutions and suggestions

Participants identified strategies to enhance their interaction with each other as a team, and with the drug development industry. To overcome the challenge of coordinating meetings that accommodate those in many different time zones, participants have highlighted the benefits of alternating meeting times each month. This has been adopted within the CAB and most members have noted an increase in effectiveness and engagement.

When reflecting upon factors for which clinical trial developers should account when developing person-centered and inclusive clinical trials, participants were most concerned with expanded access, accurately assessing drug effectiveness in different settings, and quality-of-life considerations of potential treatments. To improve industry engagement in pursuit of these goals, participants suggested continued presence in medical and pharmaceutical spaces, emphasizing the importance of attending related events and conferences to represent PLHB and D. It was suggested that CAB members could even be involved in hosting such events to provide ongoing insight and consideration into the needs and preferences of PLHB and D. Patient involvement in the planning of industry conferences has been intermittent since 1992 and has led to improved patient engagement in all aspects of healthcare and inclusion of patient-centered issues in research agendas (29).

The FDA has recently issued a "Diversity Action Plan to Improve Enrollment of Participants from Underrepresented Populations in Clinical Studies" (30). This is to enhance clinical trial diversity and help ensure that historically under-represented communities are enrolled in clinical studies. It requires drug sponsors to demonstrate that a diverse patient body was included in clinical trials for the drug. This may help to mitigate any potential power imbalance between patients and drug developers, as the industry is now mandated to consult a broad group of individuals to both inform their study design and test their products. While this does not directly reduce the possibility for conflict of interest, it is clear that the FDA is taking much-needed steps toward accountability and transparency in the clinical trial design and implementation process, and clinical trial sponsors will hopefully rise to meet these new standards.

Using well-established partnerships between patients and researchers, CAB members pinpointed strategies to create clinical trials that are inclusive, diverse, and patient-centered. Suggestions included creating easy-to-understand communication materials, encouraging communication between trial participants and researchers, and providing a human face to the research. CAB members felt that people's humanity can be lost when taking part in a study, as researchers can sometimes treat participants according to their perceived value to the study and their status as someone living with a particular health condition, rather than as whole and complex individuals (31). Collaboration between researchers and CABs can refine research outcomes by humanizing the work, uplifting patients' voices and incorporating their preferences into research outcomes (31).

The outlined strategies suggested by participants have shown positive impacts across health industries and lead to improved interactions between researchers and clinical trial participants (26, 31–33). Overall, previous research has found that when researchers engaged with advocacy organizations, their research process became more collaborative, which better incorporated the priorities of those living with the diseases of focus (34).

5 Limitations

This study has some limitations. Due to the global nature of the CAB, cultural bias may have influenced the understanding and interpretation of the focus group questions, and language barriers may have prevented focus group participants from fully expressing their thoughts. An offer of co-authorship on this manuscript was extended to all focus group participants to ensure that their views were accurately reflected.

The only geographical regions not represented in this study are South and Central America, as there are no participants from these regions in the CAB, despite persistent efforts to recruit from them. Of existing members, all 23 were offered the opportunity to participate in one of the three focus group sessions, but due to the voluntary nature of participation, seven members chose not to participate.

Social desirability bias may have impacted participants' responses. Researchers tried to mitigate this by having an external moderator lead the focus group sessions.

6 Conclusion

This CAB serves as a body of representatives for PLHB and D, with the goal of raising awareness about the challenges these groups face across the globe. This study captured the reflections of the inaugural cohort, which has been a new endeavor with efforts concentrated on establishing the CAB body's structure and building its members' knowledge and capacity. Researchers hope to repeat this process with future cohorts, the experiences of which may differ. There are lessons learned in terms of providing members the platform to effectively contribute to the design of diverse, inclusive, and patient-centered clinical trials, the development of high-quality treatments, and the possible advancement of a hepatitis B cure and more effective hepatitis delta therapeutics.

Participants felt that CAB participation has a positive impact on their lives, and they identified effective ways to support drug development, employing their unique position as people with lived experience to enhance communication between the drug development industry and the larger hepatitis B and D community. Because CAB members are trusted by their broader communities, they can serve as ambassadors, bridging the gap between scientists and PLHB and D. This could apply to both the communication of complex scientific information, and efforts to assuage some of the fears that may exist around clinical trial participation, thus reducing barriers to participation, encouraging enrollment and facilitating the development of safer and more effective treatments. Involving patients throughout the drug development process may also help build trust and acceptability of future treatments.

Data availability statement

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

Ethics statement

The studies involving humans were approved by the Heartland Institutional Review Board (IRB) (HIRB Project No. 081122-407). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

FB-J: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. BA: Writing – review & editing. JC: Writing – review & editing. AG: Writing – review & editing. YI: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Supervision, Writing – original draft, Writing – review & editing. SI: Writing – review & editing. SL: Writing – review & editing. TM: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. HM: Writing – original draft, Writing – review & editing. RN: Writing – review & editing. PO: Writing – review & editing. RR: Conceptualization, Formal analysis, Writing – review & editing. LS: Writing – review & editing. BZ: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. CC: Conceptualization, Funding acquisition, Project administration, Supervision, Writing – review & editing.

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Conflict of interest

The Hepatitis B Foundation receives public health program and research grants from BMS, GSK, Gilead Sciences and VBI Vaccines. Chari Cohen serves on a patient/advocacy advisory committee for GSK and Gilead Sciences, with funds being distributed to the Hepatitis B Foundation. Beatrice Zovich has also served on an advisory committee for Gilead for which she was financially compensated. Yasmin Ibrahim serves on a patient/advocacy advisory committee for Roche/Genentech, with funds being distributed to the Hepatitis B Foundation.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1437502/full#supplementary-material

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Disability health in medical education: development, implementation, and evaluation of a pilot curriculum at Stanford School of Medicine

Richard W. Sapp[1]*, Edmund Lee[2], Sylvia Bereknyei Merrell[2], Erika Schillinger[3], James N. Lau[2], Heidi M. Feldman[4] and Cori McClure Poffenberger[5]

[1]Stanford University School of Medicine, Stanford, CA, United States, [2]Department of Surgery, Stanford University School of Medicine, Stanford, CA, United States, [3]Division of Primary Care and Population Health, Department of Medicine, Stanford University, Stanford, CA, United States, [4]Division of Developmental-Behavioral Pediatrics, Department of Pediatrics, Stanford University School of Medicine, Stanford, CA, United States, [5]Department of Emergency Medicine, Stanford University School of Medicine, Stanford, CA, United States

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EDITED BY
María Del Carmen Valls Martínez,
University of Almeria, Spain

REVIEWED BY
Lise Mogensen,
Western Sydney University, Australia
Linda Long-Bellil,
University of Massachusetts Medical School,
United States

*CORRESPONDENCE
Richard W. Sapp
✉ rwsapp@mgh.harvard.edu

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Background: People with disabilities face significant healthcare disparities due to barriers to accessing care, negative attitudes of providers, and lack of education on disabilities for healthcare professionals. Physicians report discomfort when interacting with patients with disabilities, adding to the disparity, warranting research on medical school education.

Objective: Two educational interventions were structured: (1) a brief 2-h intervention in the mandatory curriculum and (2) a 9-week elective course which included interactions with individuals with disabilities through workshops and partner programs. We predicted that both of these interventions would result in improvements in attitude and empathy toward individuals with disabilities and reduce student anxiety.

Methods: During the 2018–2019 academic year, 54 students completed the surveys for the 2-h intervention and 8 students completed the 2-h intervention and elective course. Pre-, post-, and delayed post-intervention surveys (3 months after post survey) measured students' attitudes, using validated surveys on attitudes, empathy and anxiety toward individuals with disabilities.

Results: Both educational interventions resulted in improved attitudes toward individuals with disabilities. However, students reported only feeling prepared to care for patients with disabilities after the elective course. The elective course, but not the 2-h course, significantly decreased student anxiety levels, likely due to more individual time working with individuals with disabilities. Delayed analysis after 3 months showed that both interventions had a lasting impact on attitudes and behavior change when caring for individuals with disabilities.

Conclusion: Medical education is effective at improving medical students' attitudes and behaviors toward individuals with disabilities. A 2-h session can lead to a modest improvement in attitudes. However, more dedicated time and exposure to persons with disabilities results in a greater improvement in students' attitudes, anxiety and preparedness.

KEYWORDS

disability, attitudes, medical students, education, medical education

Background

Disability affects 61 million people in the United States, or 1 in 4 (26%) people in the total population (1). This proportion is likely to increase as new clinical treatments and public health approaches prolong the lives of individuals with chronic conditions and disabilities. A significant proportion of individuals with disabilities require multi-specialty and complex care, and have been identified as frequent healthcare utilizers (2, 3). Physicians within all specialties care for patients with disabilities, and ideally can provide treatment with comfort and competence (4). However, current evidence suggests that there are significant healthcare disparities for individuals with disabilities, resulting from structural, socioeconomic, and attitudinal barriers that contribute to both inadequate access to care and poor quality of care (5, 6). Of these barriers, people with disabilities commonly report physician attitudes as a major obstacle when engaging with the US healthcare system (7–10).

Historically, medical education has paid limited attention to issues related to healthcare for individuals with disabilities (11–13). In the absence of explicit training, students may develop negative attitudes when working with individuals with disabilities (14). On the contrary, early and frequent encounters with individuals with disabilities may improve medical students' knowledge, attitudes and skills regarding their care (15, 16). Despite a call to action for disability-based medical education from the US Institute of Medicine (17), the Office of the Surgeon General of the United States (18), and the Department of Health and Human Services (19), the lack of a curricular focus on disabilities remains the norm at many medical schools (11–13). Fortunately, curricula in disability have been developed in recent years, using a variety of methods including didactic lectures, home visits, and presentations in panels of individuals with disabilities. These methods have shown success in improving knowledge, skills and attitudes toward individuals with disabilities (20–27). Toolkits have been created to help integrate disability health into medical education and educational sessions have been created to address ableism and microaggressions (28–31). Within the past 4 years, more disability elective courses such as the one in our study have been developed and initiated by medical students (32). Additionally, disability competencies for healthcare education have recently been established by national consensus (33) and have been used to evaluate existing medical school curricula (34), finding a need for better integration of disability competency training throughout medical school education and training.

At Stanford School of Medicine in 2018–2019, we developed and implemented two novel disability-based medical education interventions to improve medical students' knowledge, attitudes and skills pertaining to patient-centered care of people with disabilities: (1) a required 2-h session during the first-year mandatory curriculum and (2) a 9-week preclinical elective course with an incorporated patient partner program. The impact of these sessions on students' knowledge, skills, and attitudes toward individuals with disabilities was measured through validated survey tools.

Methods

Educational interventions

Interventions were created with the motto "nothing about us without us in mind" (35, 36); individuals with disabilities were integrated and involved in every step of the development and implementation of the curriculum. Faculty, students, staff and individuals with disabilities worked together to create two-linked educational interventions: (1) Required 2-h session for first-year students as part of the first year Practice of Medicine (POM) course titled "Disability Health" and (2) 9-week elective course (Disability elective: Caring for Individuals with Disabilities).

"Disability health" session

To develop this session, multiple meetings were held with faculty, students, and community members who identified with and without disabilities to determine the learning objectives and design of the session. The result of these discussions was a 3-part session: a brief lecture, panel discussion, and small group case discussions. The lecture, panel questions, and cases were created and reviewed by all members of the committee until consensus was met.

The 2-h "Disability Health" session is a component of the first year Practice of Medicine (POM) curriculum, consisting of three parts: (1) a 15-min didactic session on healthcare disparities that individuals with disabilities face, the language around disability, and a comparison between the medical and social models of disability, (2) a 1-h panel with 5 local individuals with disabilities from both outside and within the school of medicine (faculty members, medical students and community members), representing different disabilities, ages, race/ethnicity, and functional strengths and needs, which focused on panelists' positive and negative experiences within the healthcare system, along with advice for improving healthcare interactions, and (3) a 40-min case-based discussion involving three cases that discuss shared decision making, non-verbal communication, and disability etiquette (Figure 1A). Cases can be found in Supplemental Appendix 1. Case facilitators included the panelists, community members with disabilities, and medical school faculty with expertise in disability health. The overall learning objectives for the session were to: (1) define disability, (2) describe the relevance of the construct of disability to the practice of medicine, (3) contrast the medical model and social model of disability, (4) identify common challenges in providing health care for individuals with disabilities and discuss strategies for improvement, and (5) develop skills for inclusive conversations around disabilities.

Disability elective

The 9-week elective course (24 h of content) (Figure 1B) was developed to expand on the major learning objectives from the

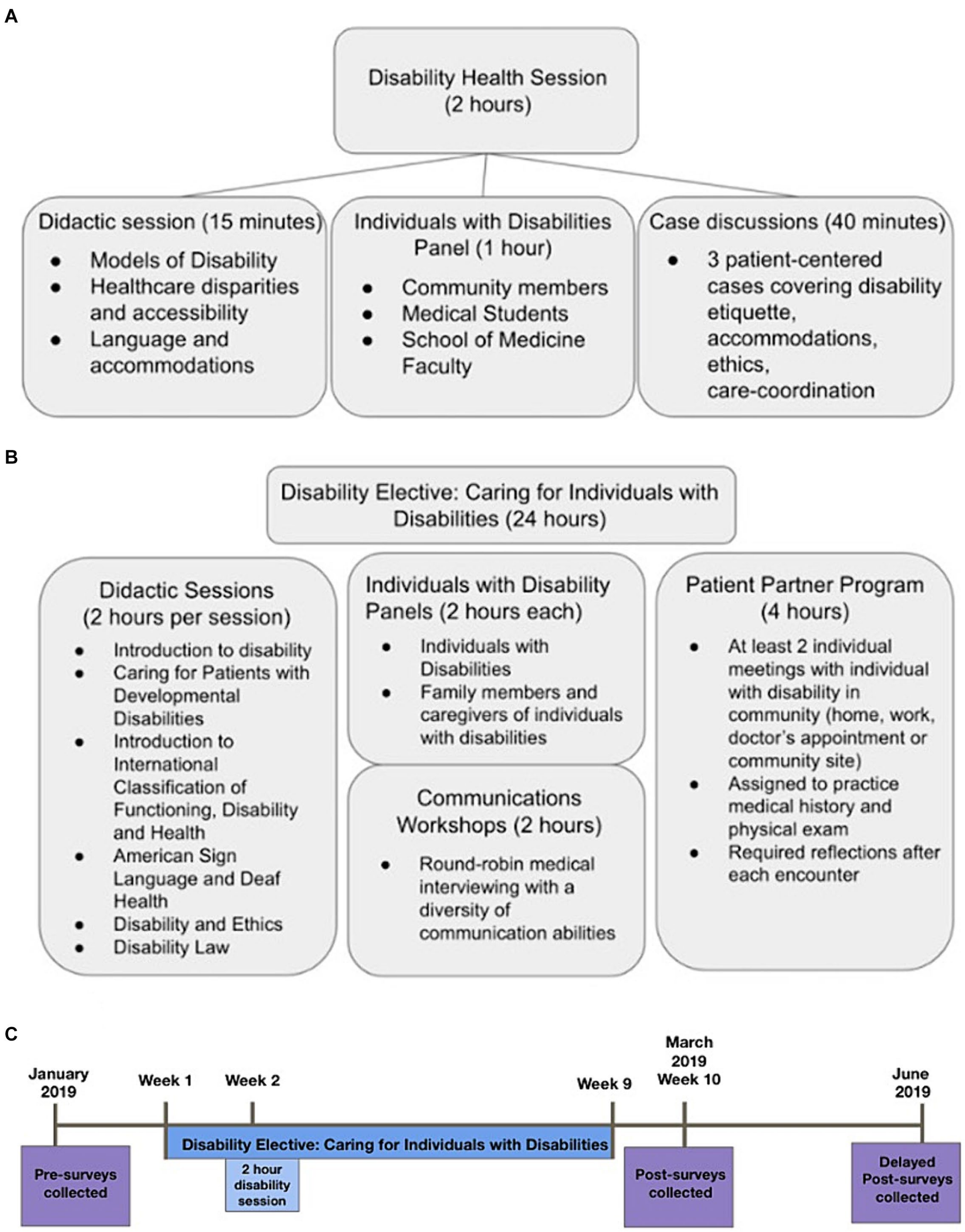

FIGURE 1
(A) Educational components of the 2 h disability health session. **(B)** Educational components of the disability elective course. **(C)** Timeline of survey administration.

Practice of Medicine session. The learning objectives were based on prior curricula (23) and from "Core Competencies on Disability for Health Care Education," which was created by the Alliance for Disability in Health Care Education (33, 37). The overarching goals of the course were to: (1) to build general knowledge of common disabilities, and to dispel misconceptions and misunderstandings, (2) to instill attitudes and commitment to patient-centered care for people with disabilities, (3) to foster skills necessary for patient-centered care for people with disabilities. We worked with community members with disabilities to design the content of the curriculum based on the above goals. The schedule of the elective was the following: Week 1—Introduction to Disability, Week 2—Individuals with Disabilities Panel, Week 3—Caregivers of Individuals with Disabilities Panel, Week 4—Caring for a Patient with a Developmental Disability, Week 5—Communications Workshop, Week 6—Disability and Ethics, Week 7—International Classification and Functioning, Week 8—Disability Law, Week 9—ASL and Deaf Health.

The classroom components included seminar discussions, panel discussions with individuals with disabilities and parents/caregivers of individuals with disabilities, and a communication workshop (round-robin 15 min mini-medical history interviews with individuals with a diversity of communication abilities). Prior to Weeks 2 and 3, panelists reviewed and provided feedback on the structured discussion prompts. Participants in the communications workshop gave direct feedback to the medical students on their interactions during history-taking and physical exam practice. The community component included a patient-partner program in which students were paired with an individual with a disability in the community. Students were required to meet with their partner at least twice in two different locations (home, school, work, doctor's office), and practice eliciting a history during one of those meetings. Individuals with disabilities provided insights about their disability experience, and students were required to write reflections after each meeting synthesizing their experiences.

Participants

This study was conducted in a single private medical school in California (Stanford School of Medicine). Eligible individuals included preclerkship medical (MD) and physician assistant (PA) students (in their first or second year of professional school) who were 18 years or older. The study was approved by the Institutional Review Board (IRB) of Stanford University (IRB-47216).

Survey content and measurement instruments

Demographics

Gender, age, ethnicity and training year were gathered at the beginning of data collection prior to the first survey. Demographic questions were taken from prior studies (38, 39). In addition, students were asked about their experiences with individuals with disabilities, including whether they self-identified as having a disability, a set of questions that had not been asked in the prior surveys.

The assessment was designed to assess the degree of change in attitudes, empathy, anxiety and competency. The survey was pretested and edited at the Goodman Surgical Education Center with experts in medical education and with individuals with disabilities who assisted in the development of the curriculum.

Attitudes

We used 2 existing questionnaires to assess attitude change as a function of education:

1. Medical Student Attitudes Toward Persons with Disabilities (MSATPD): is a 30-item questionnaire to measure medical students' attitudes toward people with disabilities (38). It comprises six factual/demographic items and 24 opinion questions. It utilizes a 4-point Likert scale ranging from 1 (Strongly Disagree) to 4 (Strongly Agree), with a higher score indicating a more positive attitude. The Total Score was the sum of scores on individual items out of 92. Item 16 ("If I introduced a person with disabilities to my friends, I think they would feel uneasy") was not analyzed due to it not loading onto any component during the psychometric content analysis, and there being a lack of pattern in participant responses (38). This instrument has been used to measure changes in medical students' attitudes in two prior studies (15, 40). The scale demonstrates good internal consistency in this study ($\alpha = 0.815$).
2. Disabilities Attitudes in Health Care (DAHC) contains 17 items that include positively and negatively worded statements that address general attitudes, cost-effectiveness, time and energy, therapeutic potential, and educational preparation of healthcare professionals caring for people with disabilities (39). It utilizes a 5-point Likert scale ranging from 1 (Strongly Disagree) to 5 (Strongly Agree), with a higher score indicating a more positive attitude. Total score out of 85. This scale has been used to measure changes in medical students' attitudes after an educational intervention (24). The scale demonstrates good internal consistency in this study ($\alpha = 0.792$).

Empathy

We relied on the Jefferson Scale of Physician Empathy Medical Student Version (JSE-S) to measure empathy among students of health professions. Participants are asked to indicate the extent of their agreement or disagreement with 20 different statements, ranging from 1 (Strongly Disagree) to 7 (Strongly Agree) (41). A higher number on the scale indicates higher agreement, which indicates higher levels of empathy (although it is noted that the JSE-S is not disability-specific). Total score out of 140. The scale demonstrates good internal consistency in this study ($\alpha = 0.871$).

Anxiety

This 12-item scale is a modified version of the intergroup anxiety scale (42) which was adapted toward individuals with disabilities (40). The response format employs a 10-point scale ranging from "Not at all" to "Extremely" on the following items: uncertain, worried, awkward, anxious, threatened, nervous, comfortable, trusting, friendly, confident, safe and at ease (the latter six are reverse scored). A lower score on this scale indicates lower levels of anxiety. Total score out of 120. The scale demonstrates good internal consistency in this study ($\alpha = 0.896$).

Additional questions

We added seven questions, adapted from a Survey Scale section "Interacting with the Disabled" and one question from the "Advocacy" scale (43). These questions were included to measure the self-reported level of competency and knowledge about interacting and working with individuals with disabilities. A group of faculty and students, experienced in medical education and survey design, modified existing questions and developed seven new questions regarding levels of confidence. Participants are asked to indicate the extent of their agreement from 1 (Strongly Disagree) to 5 (Strongly Agree). The higher score indicates higher agreement, indicative of greater competency and confidence in interacting with individuals with disabilities.

Survey administration

Our study was completed via a survey hosted by Qualtrics® (Provo, Utah, United States). We sent an email to participants with a link to the surveys, which led directly to an information page with an online consent form, followed by the measurement instruments stated above. Students developed their own personal identification codes to facilitate the pairing of responses while maintaining anonymity.

For the mandatory disability session, all first year MD and PA students were eligible for survey participation and were emailed the pre-survey in January 2019. The mandatory session took place 1 week after the pre-survey. Students received the post-survey 10 weeks after the mandatory session. The delayed post-survey was sent 20 weeks after the mandatory session (Figure 1C).

For the 9 week disability elective course, first and second year MD and PA students were eligible to participate. The same pre-survey was also sent in January 2019. First year MD and PA students enrolled in the disability elective took both the disability session and elective concurrently, and thus only filled out one survey (personal identification codes associated with the survey prevented duplicate responses). Students received the post-survey 1 week after the end of the disability elective course. The delayed post-survey was sent 12 weeks after the end of the elective course (Figure 1C).

Data analysis

Survey responses were aggregated into Microsoft® Excel (Redmond, Washington, United States) and analyzed using IBM SPSS Statistics Version 25 (Armonk, New York, US). We compared the baseline survey with the immediate and delayed post-education surveys for two groups of students: (1) those who took the 2-h required session in the Practice of Medicine Course only and (2) those who took the "Disability Health Session" and Disability elective. For analyzing the demographics, we used Chi-Square to determine differences between the groups that just took the mandatory 2-h disability session and those who took the disability session and disability elective. When analyzing pre-post intervention, and post, delayed post intervention, we used paired-T-test for the overall scores for the different measurement tools. On individual items on the surveys, we compared them using paired *t*-tests.

Results

Demographics

The total number of participants who attended the 2-h "Disability Health" session and completed all surveys was 54 (47% response rate). The total number of participants who attended the Disability elective was 8 (100% response rate). Demographic data of participants can be found in Table 1. There were noted differences in demographic data: (1) students who only took the disability health session had more professional work experience with people with disabilities ($p = 0.045$) and (2) Students who took both the disability health session and elective had more of a career interest in working with individuals with disabilities ($p = 0.034$).

There were no reported differences in the pre-survey scores for any of the scales when taking into account of the survey respondents' demographics: class year, sex, ethnicity, age, professional/work experience, volunteering, close relative or friend with a disability, self-identification with a disability, career interest or level of perceived training (data not shown).

TABLE 1 MD/PA student respondent demographic data.

	Disability health session	Disability elective
Total n (%)	54	8
MS1	38 (70)	3 (37.5)
MS2	0	5 (62.5)
PA1	15 (28)	0 (0)
No response	1 (2)	0 (0)
Sex		
Male	19 (35)	2 (25)
Female	33 (61)	6 (75)
Other	0 (0)	0 (0)
Did not disclose	2 (4)	0 (0)
Mean age (years)	24.9	26
Ethnicity		
White	26 (48)	4 (50)
Black/African American	2 (4)	0 (0)
Asian	20 (37)	4 (50)
Native Hawaiian/Pacific Islander	1 (2)	0 (0)
Hispanic/Latino	7 (13)	0 (0)
American Indian/Alaska Native	0 (0)	0 (0)
Middle Eastern/North African	2 (4)	0 (0)
Another Race/Ethnicity/Origin	0 (0)	0 (0)
Did not disclose	2 (4)	0 (0)
Professional/work experience caring for an individual with a disability		
Yes	19 (35)	0 (0)
No	35 (65)	8 (100)
Structure experiences working with people with disabilities (i.e., volunteering, teaching) aside from medical school?		
Yes	27 (50)	2 (25)
No	27 (50)	6 (75)
Friend or relative with a disability who you see at least occasionally		
Yes	34 (63)	5 (62.5)
No	20 (27)	3 (37.5)
Identify as having a disability		
Yes	7 (13)	2 (25)
No	47 (87)	6 (75)
Career interest in working with individuals with disabilities		
None	9 (16)	0 (0)
Slight	15 (28)	1 (12.5)
Moderate	21 (39)	3 (37.5)
Strong	8 (15)	4 (50)
Very strong	1 (2)	0 (0)
Level of training received in school regarding individuals with disabilities		
None	30 (56)	1 (12.5)
A little	20 (37)	7 (87.5)
Somewhat	4 (7)	0 (0)
A lot	0 (0)	0 (0)
A great deal	0 (0)	0 (0)

Disability health 2-h session

Students valued the session highly (4.1/5) and favored the patient panel (4.6/5) over the didactics (3.8/5) and cases (3.5/5). The class was successful at meeting the educational objectives for the students: (1) Define disability (3.9/5), (2) Describe the relevance of the construct of disability to the practice of medicine (4.1/5), (3) Contrast the medical model and social model of disability (4.1/5), (4) Identify common challenges in providing health care for individuals with disabilities and discuss strategies for improvement (4.2/5), and (5) Develop skills for inclusive conversations around disabilities (3.8/5). Students stated they felt they gained more awareness and a better understanding about how to talk about disability. 80% (42/54) of students were interested in further education on disability in the curriculum.

Comparing the total scores on the Pre-vs. Post-scales for the students who received the required 2-h curricular intervention demonstrated a statistically significant increase on the "Medical Student Attitudes Toward Persons with Disabilities" scale (68.91 vs. 71.19, $p = 0.0021$) (Figure 2A) but no change on the Disability Attitudes in Health Care scale (DAHC), Jefferson Scale of Empathy (JSE) or Anxiety Scale (Supplementary Figures S1A–C; Supplementary Tables S2–S4). A delayed post-test was given 3 months after the administration of the initial post-test and there was no significant change on any of the scales (Figure 2B; Supplementary Figures S1D–F; Supplementary Tables S2–S4).

When looking at the specific items of the Medical Students Attitudes Toward Persons With Disabilities Scale, there were three statements which were significantly different indicating a more positive attitude toward individuals with disabilities: "Most people with disabilities feel sorry for themselves (reversed)" (3.13 vs. 3.32, $p = 0.048$), "People with disabilities are as happy as people without disabilities" (2.96 vs. 3.2, $p = 0.05$), and "Most people with disabilities resent people without disabilities (reversed)" (3.24 vs. 3.52, $p < 0.0001$). In Part B of the scenario section of the survey where they answer questions regarding a hypothetical scenario with a man with a disability and a woman of the same age in the room to be evaluate by a healthcare provider, the following statement was significant for students "In scenario B, I would be comfortable determining the role of the man vs. the woman in providing the history of the complaint" (Supplementary Table S1).

On the individual statements of the Disabilities Attitudes in Health Care survey, Jefferson Scale of Empathy, and Anxiety scale there were no significant differences comparing the pre-and post-test surveys (Supplementary Tables S2–S4).

Students were found to have significant improvements to their confidence in their understanding of "disability" and the barriers to healthcare access, barriers to participation and quality of life issues. The following statements were statistically significant: "I am comfortable providing assistance appropriately to a person with a disability" (3.39 vs. 4.17, $p = 0.049$), "I feel confident in my understanding of 'disability'" (3.12 vs. 3.91, $p < 0.0001$), "I feel confident in knowledge of barriers to access to care for persons with disabilities" (2.7 vs. 3.56, $p < 0.0001$), "I feel confident in my understanding of cultural, economic, and physical barriers to participation" (2.7 vs. 3.4, $p < 0.0001$), and "I feel confident in knowledge about my understanding about the quality of life issues for people with disabilities" (2.6 vs. 3.41, $p < 0.0001$). It was found that there was a significant increase in the statement "I feel prepared to take care of patients with disabilities (2.07 vs. 2.48, $p = 0.014$), however, when looking at the absolute score students were reporting between neutral and disagree on this statement (2.48/5) (Supplementary Table S5).

Disability elective

Overall, the students highly rated the overall value of the disability elective (4.75/5). They rated sessions with interactions with individuals with disabilities highly: Individuals with disabilities panel (4.625/5), Caregivers of individuals with disabilities panel (4.5/5),

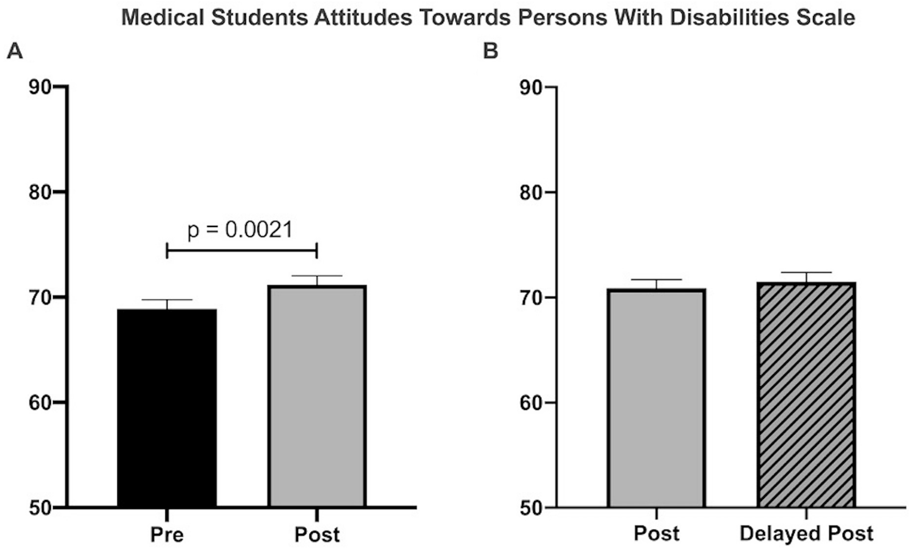

FIGURE 2
"Disability Health" 2-h session measured by the Medical Students Attitudes Toward Persons With Disabilities Scale. **(A)** There was a significant increase in attitudes toward persons with disabilities (68.9 vs. 71.2); $n = 54$, $p = 0.0021$, Students paired *t*-test. **(B)** There was no significant difference between post and delayed posttests (70.9 vs. 71.5, $n = 49$, $p = 0.42$, Students paired *t*-test).

communications workshop (4.5/5) and partner program (4.75/5). Students spent an average of 4.625 h with their partner outside of class. Didactic sessions were also rated highly: Introduction to disability (4.167/5), Caring for a patient with a developmental disability (4.167/5), Disability and Ethics 4.0/5, International classification and functioning (3.625/5), Disability law/cases (5/5), and ASL/Deaf health (4.5/5).

Students who were enrolled in the 9 week disability elective demonstrated significant improvement in attitudes and decreased anxiety when comparing pre and post-tests. On the Medical Students Attitudes Toward Persons with Disabilities scale, there was a significant improvement in attitudes (70.4 vs. 77.8; $p = 0.043$) (Figure 3A) but not on the DAHC scale (Supplementary Figure S2A). Students had significantly decreased anxiety toward individuals with disabilities based on the anxiety scale (54.63 vs. 40.13; $p < 0.1$). There was no difference in the total score of the Jefferson Scale of Empathy (Supplementary Figure S2B). Three months after the post test, a delayed posttest was given and there was no significant change on any of the scales (Figures 2D, 3B; Supplementary Figures S2C,D).

On the Medical Students Attitudes Toward Persons with Disabilities scale, there were two significant statements demonstrating a more positive attitude toward individuals with disabilities "I would be comfortable interacting with a person with an intellectual disability who was in the community on his or her own (i.e., without staff

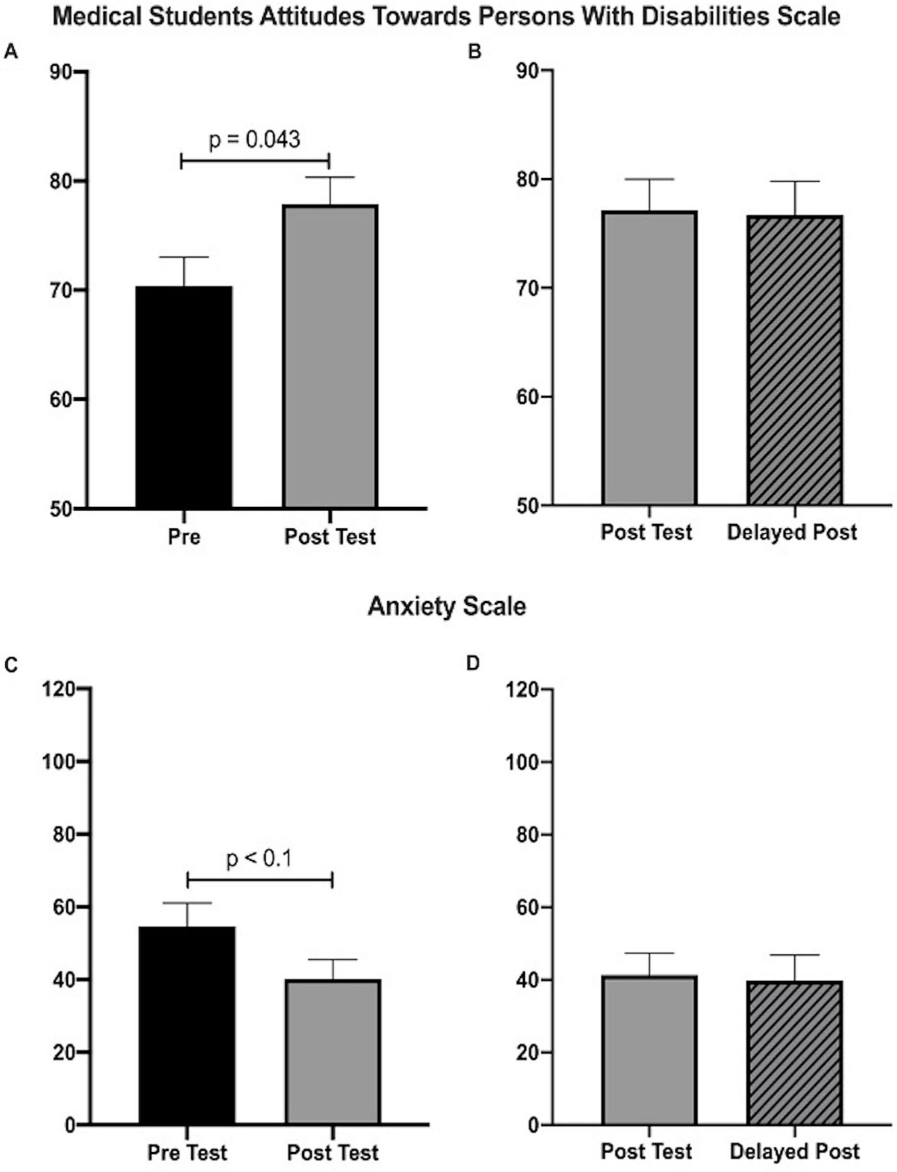

FIGURE 3
Disability Elective measured by Medical Students Attitudes Toward Persons with Disabilities Scale and Anxiety scale. **(A)** There was a significant increase in attitudes toward persons with disabilities as measured by the Medical Student Attitudes Toward Persons with Disabilities scale (70.4 vs. 77.8; $n = 8$, $p < 0.05$; Students paired t-test). **(B)** There was a marginally significant decrease in anxiety toward interacting with individuals with disabilities as measured by the Anxiety Scale (54.63 vs. 40.13; $n = 8$, $p < 0.10$; Students paired t-test). **(C,D)** There was no significant difference in post and delay post for either the Medical Students Attitudes Toward Persons with Disabilities Scale (77.14 vs. 76.85, $n = 7$, $p = 0.76$; Students paired t-test) and Anxiety scale (40.23 vs. 39.9, $n = 7$, $p = 0.53$; Students paired t-test).

members or caretakers)" (2.875 vs. 3.625, $p = 0.048$) as well as "In Scenario B, I would be comfortable determining the role of the man vs. the woman in providing the history of the complaint" where the man has a disability (2 vs. 3.625, $p < 0.018$) (Supplementary Table S6).

There were no individual items of significance on the Jefferson Scale of Empathy (Supplementary Table S8) and specific items on the Anxiety Scale trended toward significance, which included being less "nervous," "uncertain" and "worried," and being more "friendly" (Supplementary Table S9).

Students in the elective course had significant improvement on the comfort and confidence on individual items when comparing the pre and post surveys (Supplementary Table S10). In regards to the statement "I feel prepared to take care of patients with disabilities," students felt more prepared after the disability elective (2 vs. 3.625, $p = 0.006$, Supplementary Table S10). In addition, students scored significantly higher on their understanding of patients with disabilities and their knowledge on barriers to access of care: "I feel confident in my understanding of 'disability'" (3.25 vs. 4.13, $p = 0.04$), "I feel confident in knowledge of barriers to access to care for persons with disabilities" (2.38 vs. 4.38, $p = 0.01$), and "I feel confident in my understanding of cultural, economic, and physical barriers to participation" (2.88 vs. 4.50, $p = 0.02$). On survey items related to comfort in performing aspects of a history and physical exam with a patient with a disability, students performed significantly higher on the following statements: "I am comfortable adapting my body positions to make someone who uses a wheelchair more comfortable" (2.88 vs. 4.25, $p = 0.01$), "I am comfortable adapting my body positions to facilitate effective communication for someone who is visually or hearing impaired" (3.63 vs. 4.75, $p = 0.03$), "I am comfortable adapting my interviewing technique to accommodate patients with disabilities" (2.75 vs. 4.25; $p = 0.01$) and "I feel confident in communicating with patients with disabilities" (2.63 vs. 4.25, $p = 0.02$).

Comparison of educational interventions (disability health session vs. disability elective)

In comparing the baseline pre-survey scores between both educational interventions, there was no statistical difference on any of the survey instruments (Table 2).

Overall, both interventions resulted in improvement in students' attitudes toward individuals with disabilities. When comparing the students who received just the 2 h Disability Health session, with those who also took the Disability Elective course, there was a significant difference in post survey attitudes as reflected by the Medical Students Attitudes Toward Persons with Disabilities Scale (71.2 vs. 77.8, $p = 0.006$) (Figure 4A). In addition, anxiety was significantly reduced for students in the Disability elective compared to the students who only took the 2 h disability health session (54.8 vs. 40.13, $p = 0.007$) (Figure 4B). The differences in attitudes and anxiety were maintained on a repeat survey 3 months after the educational interventions were completed (Supplementary Figure S3A,B).

When looking at the individualized statements from the surveys, students who took the elective class had much less overall anxiety when caring for patients with disabilities (less uncertain, less worried, less threatened, less anxious and more at ease, more comfortable and safe; Supplementary Table S14). Students who took the elective course compared to students taking just the 2-h session felt more "comfortable adapting my body positions to facilitate effective communication for someone who is visually or hearing impaired" (4.11vs. 4.75, $p = 0.045$), more "confident in communicating with patients with disabilities" (3.26 vs. 4.25, $p = 0.017$), and more "confident in my understanding of cultural, economic and physical barriers to participation" (3.44 vs. 4.5, $p = 0.007$) (Supplementary Table S15). Students overall in the disability elective felt more "prepared to take care of patients with disabilities" (2.48 vs. 3.625, $p = 0.005$).

TABLE 2 Data table summarizing the measurement instruments across the educational interventions.

	Pre-survey	Post-survey	Pre- vs. Post	Delayed post-survey	Post vs. Delayed post
Medical student attitudes toward persons with disabilities (MSATPD)					
Disability session ($n = 54$)	68.9	71.2	**$p = 0.0021$**	71.5	$p = 0.42$
Elective course ($n = 8$)	70.4	77.8	**$p = 0.043$**	76.85	$p = 0.76$
Session vs. Elective	$p = 0.28$	**$p = 0.006$**		**$p = 0.05$**	
Disabilities attitudes in health care (DHCA)					
Disability session ($n = 54$)	68.9	68.9	$p = 0.99$	68.5	$p = 0.57$
Elective course ($n = 8$)	70.25	69.5	$p = 0.75$	71.4	$p = 0.51$
Session vs. Elective	$p = 0.36$	$p = 0.8$			
Jefferson scale of empathy (JSE-S)					
Disability session ($n = 54$)	120.3	120.3	$p = 0.97$	121.3	$p = 0.38$
Elective course ($n = 8$)	123.25	122.3	$p = 0.63$	125.3	$p = 0.17$
Session vs. Elective	$p = 0.31$	$p = 0.63$			
Anxiety Scale					
Disability session ($n = 54$)	56.1	54.8	$p = 0.35$	54.8	$p = 0.95$
Elective course ($n = 8$)	54.63	40.13	**$p = 0.1$**	39.9	$p = 0.53$
Session vs. Elective	$p = 0.64$	**$p = 0.0007$**		**$p = 0.01$**	

Bolded p-values indicate statistical significance.

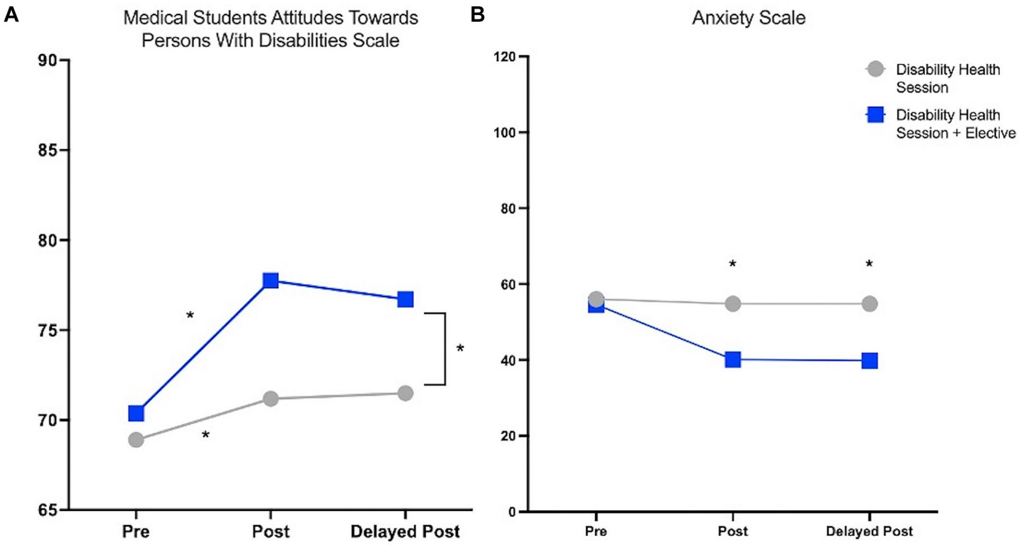

FIGURE 4
Comparing the Post Survey evaluations between the Disability health 2 h session versus Disability Elective measured by Medical Students Attitudes Toward Persons with Disabilities Scale and Anxiety scale. **(A)** Students who took the Disability health session and Disability elective had significantly higher scores on the Medical Students Attitudes Toward Persons with Disabilities scale (71.2 vs. 77.8, $p = 0.006$; Student's t-test). Delayed post survey shows that the scores were maintained 3 months after the post survey was completed (71.5 vs. 76.9, $p = 0.05$). **(B)** Students who took the Disability health session and Disability elective had significantly lower scores on the Anxiety scale indicating lower anxiety (54.8 vs. 40.13, $p = 0.0007$; Student's t-test). Delayed post-survey shows that the scores were maintained on the Anxiety scale 3 months after the course was completed (54.8 vs. 39.9, $p = 0.01$; Student's t-test).

Discussion

People with disabilities represent 26% of patients in the US, and yet many physicians feel inadequately equipped to care for this population. As a result, several national organizations have published calls to action to improve medical education surrounding caring for those with disabilities (21, 27). In this study, our objective was to evaluate the impact of two curricular interventions on medical students' attitudes, empathy, and anxiety when caring for individuals with disabilities.

Our study demonstrated that the mandatory disability session improved student attitudes toward individuals with disabilities based on the Medical Student Attitudes Toward Persons with Disabilities survey (15). However, the total score increase was modest. While students reported that they felt more prepared after the mandatory disability session, the absolute values of the preparedness scores still showed that the average students still felt neutral or disagreed that they were prepared to care for patients with disabilities. The survey response indicates that while a 2 h session can improve self-reported preparedness, it is not sufficient for medical education on disability health. Importantly, although other disability education studies have not investigated the long term impact of interventions, our study noted that improved attitudes remained at the time of the delayed post-survey, 3 months after the intervention (27). Although our study showed there was benefit 3 months after the intervention, a recent study has shown that in another disability curricular intervention attitude and empathy gains 1 year later did not sustain the benefits (44). The 2 h curriculum did not reduce student's anxiety levels toward caring for individuals with disabilities, which we hypothesize was due to a lack of direct interaction with this population. In both interventions, the students did not score differently on the Jefferson Scale of Empathy. One possible explanation is that the survey is not disability specific. Another potential rationale for this finding is that medical students in our study had a high baseline empathy score (mean = 120.3) compared to the JSE mean when it was created (mean = 114.3). This difference in baseline empathy scores may result from prior exposure to other modules in medical school curricula and through screening through the medical school's admissions process, and as a result our intervention may have had a lower potential impact on JSE scores. In contrast to their improvement in attitudes on the Medical Student Attitudes Toward Persons with Disabilities survey in both intervention groups, there was no difference on the Disabilities Attitudes in Health Care scale. This difference could be due to the way the two tools were developed. The DAHC was adapted from two prior scales which were based on geriatric scales (39), whereas the MSATP was adapted from seven scales and incorporated significant input from the disability community including patients and families, medical educators, and local professionals who work with people with disabilities (15, 45). Although students' attitudes and preparedness improved on the MSATP with the 2 h curricular intervention, the majority of students still disagreed with the statement that they felt prepared to care for patients with disabilities, which indicates although there was a statistical increase, there was likely a gap in clinical significance, arguing for the importance of more spaced repetition and integration of disability health throughout the longitudinal medical curriculum. When developing the disability elective course, we specifically designed it to include more direct interaction with individuals with disabilities and skills training surrounding the recommended core competencies with the hypothesis that it would have greater impact on attitudes and behavior (13, 37).

We demonstrate here that the 9 week elective course was an effective educational intervention. The students who took the elective course scored significantly higher on the Medical Students Attitudes Toward Persons with Disabilities Scale and lower on the Anxiety scale compared to the 2 h disability session alone. Their reduction on the Anxiety scale supports the intergroup contact hypothesis, where frequent and early interactions with individuals with disabilities in medical training improves comfort (15, 46–48). After the communications skills workshops, students felt more confident in communicating with patients with disabilities compared to their counterparts who only did the 2 h session. The ability to practice skills and receive feedback likely contributed to overall feeling prepared. Students who took the comprehensive elective course on average agreed that they were prepared to care for individuals with disabilities, unlike the students who just took the 2 h session.

Given the positive impact of our curriculum, we hope that all medical schools consider adopting similar sessions to improve disability competency. We show here that even a short 2-h disability session can improve medical student attitudes toward patients with disabilities, however, is not sufficient to help students feel prepared. The elective could be taught as a stand-alone course as is the trend among other medical schools with the development of disability health electives (32) or components of our elective session could be included throughout required medical school training (23) and be utilized to modify core EPAs to cover competencies (13). Further research would include incorporating components of the disability elective curriculum into the mandatory curriculum and evaluating students throughout different stages of their medical education.

Given the challenge of limited time to add additional sessions into medical school curricula, we propose ways to include components of our elective course into existing parts of standard medical school didactics. Didactic sessions from the elective which include the history of disability and the ethics of disability, and teaching on the social model of disability could be converted into online modules for ease of student accessibility to the information. Additionally, during practicum skills sessions, individuals with disabilities should be included during history and physical exam teaching sessions, as it has been shown that students do not perform as well on practical skills exams with individuals with disabilities if they have not received specific practice prior (49). Educators at medical schools have expressed difficulty in incorporating people with disabilities as teachers, however, we found in development of our course that there are many existing local and national organizations that medical schools can partner with to gain access to disability educators.

Our study differs from other published brief disability curricular interventions in that we uniquely utilized members of the healthcare professions with disabilities to participate in the patient panel and in the discussion groups, in order to normalize disability as diversity in the medical profession. A study at Stanford that showed 28.4% of faculty, students, and staff in the School of Medicine reported having an ADA defined disability. However, public self-identification of disability within the institution is rare for individuals with invisible disabilities (50). Our educational interventions also included participants with a wide range of disabilities, showcasing the diversity within disability.

This study was not without significant limitations. Overall, the study would have vastly benefited from a greater sample size, as the numbers in this study were very modest, and randomization of participants into the different interventions, to try and reduce the bias of students with greater interest taking the elective course. The study design would have strongly benefited from a control group from a different institution who did not have any disability curriculum at the time of intervention. Since students chose to take the longer elective course, there could be bias between the two groups in their motivation to learn about patients with disabilities. People in the elective course had more career interest in working with individuals with disabilities, which could explain the trend of slightly more positive attitudes on the pre-survey MSATPD scale (70.4) compared to the other group (68.4), although this was not statistically significant. Low response rates and participant retention is a complication of web-based data collection, and the sample of participants in this study may not be entirely representative of the general medical student population. Generalizability of the finding is also limited by the location of the study at only one medical school. Additionally, we recognize that survey measures used in this study are not direct proxies for providers' actions. In the future, studies comparing students' self-rated scores to patient's perceptions of their care, and overall patient outcomes would be more direct measures of the effect of our interventions. In light of limitations, the study strengths included a multi-disciplinary team approach from multiple perspectives in medicine. Additionally, our surveys were anonymous, minimizing social desirability bias.

Conclusion

Our findings provide support that both a brief 2 h curricular intervention and a more comprehensive 9 week elective curriculum can improve medical students' attitudes toward individuals with disabilities with a long term effect. In comparing the two interventions, we highlight the importance of direct interaction with individuals with disabilities as specific communications workshops, panels, and partner programs further reduced anxiety and better prepared medical students to care for individuals with disabilities. We hope that similar curriculums can be incorporated at medical schools across the country. Reducing healthcare disparities toward individuals with disabilities will require a multifaceted approach with system wide changes in our healthcare system, and it is essential that medical students have exposure to individuals with disabilities as a basic tenet to improve healthcare for this population.

Data availability statement

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

Ethics statement

The studies involving humans were approved by Institutional Review Board (IRB) of Stanford University (IRB-47216). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

RS: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Writing – original draft, Writing – review & editing. EL: Writing – review & editing. SB: Formal analysis, Supervision, Writing – review & editing. ES: Supervision, Writing – review & editing. JL: Funding acquisition, Supervision, Writing – review & editing. HF: Conceptualization, Funding acquisition, Investigation, Supervision, Writing – review & editing. CP: Conceptualization, Investigation, Supervision, Writing – review & editing.

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Conflict of interest

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

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fmed.2024.1355473/full#supplementary-material

SUPPLEMENTARY DATA SHEET 1
Supplemental Figures and Tables.

SUPPLEMENTARY DATA SHEET 2
Supplemental Appendix 1.

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Intersectional equity in Brazil's remote rural municipalities: the road to efficiency and effectiveness in local health systems

Simone Schenkman* and Aylene Bousquat

Department of Politics, Management and Health, Faculty of Public Health, University of São Paulo, São Paulo, Brazil

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EDITED BY
María Del Carmen Valls Martínez,
University of Almeria, Spain

REVIEWED BY
Brahim Zaadoud,
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Ricardo De Moraes E. Soares,
Instituto Politecnico de Setubal (IPS), Portugal

*CORRESPONDENCE
Simone Schenkman
✉ simoneschenkman@usp.br

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Objective: The Brazilian remote rurality has been classified more reliably only recently, according to demographic density, proportion of urban population, and accessibility to urban centers. It comprises 5.8% of the municipalities, in nearly half of the states, with a population of 3,524,597 (1.85%). Remote rural localities (RRL) have reduced political/economic power, facing greater distances and barriers. Most health strategies are developed with the urban space in mind. We aim to understand how RRL are positioned concerning efficiency/effectiveness in health, compared to other urban-rural typologies of Brazilian localities, focusing on Primary Health Care (PHC), and its organizational models.

Methods: We evaluated the efficiency and effectiveness of the organizational models using the health production model, from 2010–2019, gradually deepening the immersion into the RRL reality. We analyzed the human and financial resources dimensions, emphasizing teams, the results of PHC actions, and health levels. We used the fixed effects model and data envelopment analysis, cross-sectioned by intersectional inequities. We compared the Brazilian states with and without RRL, Brazilian municipalities according to rural-urban typologies, and RRL clusters.

Results: Brazilian RRL states show superior resource/health efficiency through services utilization according to health needs. The remote rural typology demonstrated greater efficiency and effectiveness in health than the other typologies in the RRL states. The organizational models with the Family Health Strategy (FHS) teams and the Community Health Worker (CHW) visits played a key role, together with local *per capita* health expenditures and intergovernmental transfers. Thus, financial resources and health professionals are essential to achieve efficient/effective results in health services. Among the RRL, the Amazon region clusters stand out, denoting the importance of riverine and fluvial health teams, the proportion of diagnostic/treatment units in addition to the proportion of illiteracy and adolescent mothers along with the inequity of reaching high levels of schooling between gender/ethnicity.

Conclusion: Hopefully, these elements might contribute to gains in efficiency and effectiveness, prioritizing the allocation of financial/human resources, mobile FHS teams, availability of local diagnosis/treatment, and basic sanitation. Finally, one should aim for equity of gender/ethnicity in income and education and, above all, of place, perceived in its entirety.

KEYWORDS

equity, rural health, primary health care, intersectionality, efficiency, effectiveness, data envelopment analysis, fixed effects models

1 Introduction

Remote rural localities should be acknowledged according to the dynamics of their underlying socio-spatial relations, their social structures, and power relations, whose constant disputes bring material and symbolic consequences, even influencing how a remote rural space is conceived and valued locally and globally (1).

In Brazil, municipalities with these characteristics were classified more reliably to their reality recently by the National Institute of Geography and Statistics-IBGE (2), according to demographic density, proportion of urban population, and accessibility to urban centers. These localities are the rural remote municipalities ($n = 323$) of 13 Brazilian states (out of a total of 27 units), adding up to 5.8% of the municipalities, with a population of 3,524,597 (1.85%), differentiated from the other urban-rural typologies (urban, the adjacent rural and the intermediate remote or adjacent).

Moreover, remote rural localities differ from rural areas, with smaller and more cohesive populations but with reduced power, longer distances, and social and geographical barriers (3). These localities are not homogeneous; neither are their populations, which have multiple compositions and activities, far from performing only activities in the countryside (4, 5).

Furthermore, most public policies are still formulated based on the urban space; thus, decision-makers analyze the rural space in opposition to the urban space in a decontextualized way. The specific populations of these locations, especially the more remote ones, are even less considered in policy formulation, making it hard to implement them in remote rural locations because they deal with homogeneous policies focused on the urban space and its populations. These populations present greater social vulnerability and constitute the traditional peoples of the fields, waters, and forests, seldom studied (5).

As a counterpoint, we have specific studies and analyses on the health policies of Primary Health Care (PHC), with the research group *PHC in networks* dedicating itself to the theme; since the mid-2000s, bringing together researchers from various Brazilian educational and research institutions. More recently, the project "PHC in remote rural territories" (2018) seeks to recognize the characteristics of the supply of primary care services and their relationship with the network to ensure comprehensive and integrated care, aiming for local solutions to improve healthcare access and quality (5).

In this line, the best healthcare model is one whose logic, in the technical dimension of work processes and health practices, combines the best techniques and technologies to solve health problems according to the health needs of the population (6, 7), in an equitable, effective and efficient way. PHC, mainly through the Family Health Strategy (FHS), offers the best response to these needs, especially in remote rural settings that are distant from large urban centers and have high population rarefactions.

Multiprofessional PHC teams are expected to connect with families in the territory under their responsibility, offer and integrate individual and collective actions, provide the first contact, take responsibility for referral to other points in the system, navigation through health services, and continued care of patients, especially with health surveillance actions and the efforts of Community Health Workers (CHW) (8). We have 51,369 FHS teams (80% PHC coverage; 2023) and 257,061 CHW (61% coverage; 2020).

More importantly, the model should allow access and accessibility, either by facilitating the traveling needs of the population to the PHC facility, through institutional transportation, or in a virtual way with different means of communication—telehealth (9)—or even by moving the units and or health professionals towards the population that lives farther from the headquarters, such as those that live by the riverside or inside the forests (10). Their main contact with health services is through CHW home visits and with the health professionals that eventually organize local activities. Thus, fluvial and riverine teams designed especially for their needs are essential in this outreach, as well as fluvial PHC facilities that overcome distances and short local supplies and other means of transportation, such as ambulances and speedboats.

From the perspective of proper (effective) PHC, an integrative international review conducted by Franco et al. (11), defines three basic categories to delineate strategies adapted to rural and remote localities: access, health organization, and workforce. Access related to geographic aspects, users' travel needs, and access to more complex services; the healthcare organization highlighted the operation of health services and community management, the physical structure, and critical supplies. Concerning the health workforce, the professional profile, role, and the factors of attraction and retainment stood out. The authors highlighted the importance of these cross-cutting axes: community action, extension and visitation models, information and communication technologies, access to care, and training and professional development.

Added to these, other relevant dimensions to PHC outlined by de Almeida et al. (12), which discuss the organization of PHC practices, being more successful, the greater its proximity and centrality. The authors advise us to consider the adscription of the clientele and territorialization, the organization of work processes, the first contact of the service, and interprofessional work. The critical points refer to

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Abbreviations: CHW, Community Health Workers; DEA, Data Envelopment Analysis; FE, Fixed Effects Model; FHS, Family Health Strategy; FU, Federative Units; GDP, Gross Domestic Product; IBGE, Brazilian Institute of Geography and Statistics; IM, Infant Mortality rates; LEB, Life Expectancy at Birth; MATOPIBA, Maranhão, Piauí, Tocantins and Bahia; MWV, Midwest Vector; NMG, Northern Minas Gerais; PHC, Primary Health Care; PNS, National Health Survey; RRL, Rural Remote Locations; Brazilian states; AC, Acre; AL, Alagoas; AP, Amapá; AM, Amazonas; BA, Bahia; CE, Ceará; DF, Federal District; ES, Espírito Santo; GO, Goiás; MA, Maranhão; MT, Mato Grosso; MS, Mato Grosso do Sul; PA, Pará; PB, Paraíba; PE, Pernambuco; PI, Piauí; PR, Paraná; MG, Minas Gerais; RJ, Rio de Janeiro; RN, Rio Grande do Norte; RO, Rondônia; RS, Rio Grande do Sul; RR, Roraima; SC, Santa Catarina; SE, Sergipe; SP, São Paulo; TO, Tocantins.

distances and involve the availability of health transportation, information, and communication technologies, and strategies to attract and retain professionals.

1.1 Literature review: a brief overview

Despite the growing production of knowledge in these locations, most health strategies are still developed with the urban space in mind, ignoring rural locations and their specific populations, having to adapt them alone, without the corresponding resources. On another note, there are few studies that measure efficiency in PHC (13, 14), and even rarer are those that consider the inequities and local health systems of remote rural spaces. Some qualitative studies mention difficulties in allocative efficiency when showing disparities and imbalances between resources, especially human and logistic, when compared to urban settings (15). As a consequence, patients have poor access to healthcare, especially to higher-level facilities.

This trend is also observed in international settings with remote rurality, where health inequality is a global issue. For instance, Zhu et al. (16) describe how Asian-Pacific countries are having difficulties in retaining human resources in rural remote contexts, due to better wage offers in the private sector, located in urban settings. The incentives for attracting and retaining health professionals should include education, financial and personal support, with a strong PHC guidance and referral system.

In Canada, Wong and Regan (17) studied patient perspectives on PHC in rural communities, finding that the main problems were difficulties in access, continuity of care, and efficiency of the health system and services, specifically the diagnostic and therapeutic services. The traveling needs and expenses were frequently reported as a burden due to lack of organization and resources. Mseke et al. (18) conducted a scoping review in OECD countries and confirmed the impact of distances on healthcare access; mainly the time spent traveling seeking specialist medical care.

Kontodimopoulos et al. (19) actually measured productive efficiency in Greece for hospital health centers providing primary and secondary care in remote rural localities. Their findings suggest that services providing preventive medicine were more efficient regarding service utilization and hospital admissions. Also, they ponder on the needs to balance equity and efficiency in these settings. Likewise, Shen et al. (20) describe the need to enhance the quality and scope of PHC outpatient visits in remote localities, considering the health service utilization disparities in western China, where residents are provided with more medical services close to their homes. Both studies suggest that telemedicine and PHC may increase efficiency in these localities.

Mitton et al. (21) performed a broad review on the innovations on health service organization and delivery in the northern Arctic remote rural regions. Their main findings related to organizational structure of health services; utilization of telehealth and e-health; medical transportation; and public health challenges. The initiatives included operational efficiency and integration, access to care, organizational structure, public health, continuing education and workforce composition, which may positively impact health care quality and outcomes.

Regarding access and quality of care, Ferreira et al. (22) found evidence in Portuguese public hospitals that technical efficiency is predicted by access to health care services, patients' clinical safety, appropriateness and timeliness. Quality and access work in the same direction as efficiency, especially with the association between primary and secondary health care. Ferreira et al. (23) also analyzed multiple criteria for satisfaction in a Portuguese pediatric inpatient service. Although it relates to the secondary level of care, some aspects are essential in PHC as well, such as the health professional's communication skills, clinically and concerning the capacity to explain the users' rights and duties, as well as the auxiliary staff's efficiency and concern, such as the CHW.

Nunes and Ferreira (24) evaluated the efficiency and effectiveness of health before and during the COVID-19 pandemic: although efficiency dropped in the beginning of the pandemic, the effectiveness increased during the pandemic, demonstrating the sustainability and resilience of health services and professionals, and the importance of collective actions. Ferreira et al. (25) studied an optimal model for increasing the allocative efficiency of Portuguese public hospitals, according to the different modes of funding and contracts, which may tackle the inequities on access to health care and improve the quality of services, with better outcomes.

Newberry and Mallete (26) presented a pilot project that could impact health outcomes and patient experience in Canada, with Rural Health Hubs and Patient Medical Home concepts that intertwine improved patient experiences/navigation, population outcomes and system efficiency. In short, most of the international literature complies with the need for efficiency, equity or both, but few of them measure it and when they do, they do not evaluate this balance in the model, nor do they perform efficiency analysis of local health systems. On the contrary, they all outline the importance of formulating health policies according to sociogeographical characteristics, imbalances between resources, especially human and logistic, when compared to urban settings (15). As a consequence, patients have poor access to healthcare, especially to higher-level facilities.

From the brief literature review around the globe, we may affirm that place constitutes a relevant intersectional category, with urban dominance and forms of oppression in rural social space. This category adds to the other intersectional categories (27), such as gender, ethnicity, and social position, accumulating inequities. The complex notion of place has attracted much attention from geographers who describe it as a bounded entity containing unique characteristics within which people shape deep connections and identities (28). This description means that place is doubly constructed, physically but socially interpreted, narrated, perceived, felt, understood, and imagined. Accordingly, socio-environmental space is inseparable, operating simultaneously with the systems of objects and actions, time and totality (29).

Thus, places are not just physical constructs but are steep in social aspects. While places have unique meanings for people, personal history and experiences will influence their perceptions and experiences about places; at the same time, places will affect their opportunities and activities. Therefore, places interconnect in complex and unequal ways through social power relations (28).

Intersectionality encourages critical reflection that allows researchers and decision-makers to move beyond the singular categories that are typically favored in policy analysis to consider the complex relationships and interactions between gender, ethnicity, and social class, in addition to other social situations and identities, such as Indigeneity, sexuality, gender expression, immigration status, age, ability, and religion. This framework allows for examining the simultaneous impact and resistance to systems and structures of oppression and domination, such as racism, classism, sexism, ableism, and heterosexism. Thus, intersectional equity points to improving the living conditions and health status of different categories in an interrelated way (30). Achieving intersectional equity means reducing and ultimately eliminating disparities in health and its determinants that harm and affect excluded or marginalized groups (31).

To better understand the intersectional inequities in these localities and verify the best organizational models to reduce them, we aim to analyze the efficiency and effectiveness of the remote rural local health systems compared to other typologies of Brazilian localities. Thus, we intend to provide elements that can contribute to planning health actions and programs that serve these social groups in their territory, reducing the profound inequities of Brazilian society.

2 Methods

We evaluated the efficiency and effectiveness of different organizational models (arrangements of PHC teams and establishments) using the health production model (32) in remote rural socio-environmental spaces. We assessed the dimensions of human, material, and financial resources, emphasizing teams, the results of PHC health actions, and the final results (variables detailed in Table 1). The methodology used was the Fixed Effects (FE) model and Data Envelopment Analysis (DEA) (32), with the dimension of intersectional inequalities transversely evaluated.

We also present Figure 1 to make it easier to understand the analysis of the health production process, with the main variables analyzed according to the stages, from inputs/resources through health outputs to final results (outcomes), cross-cut by intersectoral variables.

Considering the few efficiency analyses at the local level, we carried out local efficiency analyses in remote rural municipalities, simultaneously comparing them to the Federative Units (FU) (states) to which they belong. At that level, it is possible to evaluate the use of health services according to health needs, which allows for overcoming problems already perceived in the local analyses. Compared to the global efficiency analyses, local analyses usually present lower determination coefficients in the health dimension (32).

We focused on equity in the financing and distribution of health teams, which involves allocating human and financial resources according to health needs and the proportion of the state's GDP devoted to health. We evaluated the following variables: sources of funding, local expenditures, and revenues for health, the proportion of allocation of resources to primary care of total health spending, staffing and funding arrangements, and transfers between different levels of government.

Regarding equity in access, we assessed the following aspects: the use of health services according to needs; financial and non-financial barriers (access, use, coverage, and prevention); the scope of actions, services, and practices, in addition to hospitalizations from PHC and basic sanitation sensitive causes.

Environmental or intersectoral variables include demographic, socioeconomic, governance, and risk factors (environmental and chronic diseases) dimensions.

2.1 Statistical analysis

We used two complementary techniques: Data Envelopment Analysis allows analysis throughout the stages of the production process (33), while the Fixed Effects model enables a dual analysis (34), combining effectiveness (results of the coefficients obtained) with efficiency (residual analysis). The number required for the sample is at least 10 observations per independent variable for the regression (FE), or $N > 104 + m$, where "m" is the number of independent variables (35). Considering that the set has approximately 300 remote rural localities (RRL) per period and we did not analyze more than twenty variables simultaneously, we have complied with the proposed rules. In the case of DEA, the rule proposed by Cooper et al. (36) is that the sample exceeds the number of inputs (m) and outputs (s) by several times, specifically that "n" is greater than max. [m*s, 3*($m + s$)]. For more details, please consult Supplementary material S1.

We performed three analyses to deepen our understanding of the remote rural localities. We have prepared a step-by-step infographic to help readers understand the immersion in the remote rural reality (Figure 2).

Our hypothesis is that RLL might have higher efficiency and effectiveness in health, considering the limited resources and their capacity to mobilize health resources, especially concerning the healthcare FHS models. We also knew of the heterogeneity of these regions and clusters of the RRL. Hence, we supposed that the collective PHC models would be stronger in distant regions, where there are mobile health facilities and customized riverine health teams.

We have tested our hypothesis in three layers of statistical analysis, in order to deepen the RRL reality. First, we studied all of Brazil's states, comparing states with and without these localities, through fixed effects models, globally and dimensionally (resources, health and intersectoral variables). The use of health services according to health needs was a very important test, since we had past experiences that the health dimension yielded low determination coefficients.

Then, we examined the RRL states and performed DEA and fixed effects, comparing the urban–rural typology. Finally, we compared different sociogeographical clusters within the RRL.

2.2 Empirical strategy

Our empirical strategy comprises the use of logistic regression in order to assess the utilization of services according to health needs, the fixed effects model and data envelopment analysis.

Fists, we explored health and sociodemographic variables to measure service utilization with a logistic regression model in all Brazilian states. The advantages of performing logistic regression is the small number of requirements, such as not presenting multicolinearity nor heterocedasticity, along with high reliability. Maximum likelihood is used to estimate the parameters., presented either by its linear form, *logit* (P) or the probability odds log ($p/(1 - \mathrm{p})$), with P exhibiting the formulae:

$$P(y=1)=\frac{1}{1+e^{-f(x)}}$$

TABLE 1 Variables in national comparisons, for Brazilian states and remote rural localities, according to the stage of the health production process, data sources and periods analyzed.

Dimensions	Variables	Period 1 (t)	Period 2 ($t+1$)
Input			
Financial resources	Total health expenditure *per capita*	2010 IBGE	2019 STN/MF
	GDP composition	2010 IBGE	2019 STN/MF
	Public finance (taxes and transfers)	2010 IBGE	2019 STN/MF
	Health spending as a % of GDP	2010 IBGE	2019 STN/MF
	Primary health care (PHC) expenditure	2010 SIOPS/MS e-gestor-AB	2019 SIOPS/MS e-gestor-AB
Human, material and technological resources	Density of health professionals	2010 CNES	2019 CNES
	Number of PHC doctors and nurses	2010 CNES	2019 CNES
	Number and density of PHC facilities	2010 CNES	2019 CNES
	Number and density of other services and mobile units	2010 CNES	2019 CNES
	FHS (types of teams) and CHW coverage	2010 CNES e-gestor-AB	2019 CNES e-gestor-AB
	Density of hospital beds/equipment	2010 CNES	2019 CNES
Output			
Access and use	Consultations (per inhabitants)	2010 SAI	2019 SAI
	PHC consultations/home visits	2010 SIAB	2019 SISAB
	Hospital admissions (per 100 inhabitants)	2010 SIH	2019 SIH
	Prenatal consultations—consultation coverage	2010 SINASC	2019 SINASC
Prevention	Incidence of tuberculosis and HIV	2010 SINAN	2019 SINAN
	Vaccination—vaccination coverage	2010 PNI	2019 PNI
	Cervical cancer screening	2010 SISCOLO	2019 SISCOLO
	Breast cancer screening	2010 SISMAMA	2019 SISMAMA
*PHC (intermediate results)	% of adult mothers and newborns with normal weight*	2010 SINASC	2019 SINASC
	Hospitalizations for PHC and basic sanitation sensitive conditions*	2010 SIA/SIH	2019 SIA/SIH
	Relationship between use and need	2013 PNS	2019 PNS
Outcomes			
Final results	Life expectancy at birth	2010 IBGE	2016 IBGE
	Infant mortality	2010 SIM/SINASC	2016 SIM SINASC
	Deaths—preventable causes	2010 SIM	2015 SIM
Environmental/intersectoral			
Demographic and socioeconomic	Fertility rate	2010 IBGE	2019 IBGE
	Dependency ratio	2010 IBGE	2019 IBGE
	Population density	2010 IBGE	2019 IBGE
	Gini index of *per capita* household income	2010 IBGE	2019 IBGE
	% Bolsa Família (cash transfers)	2010 MDS	2019 MDS
	Municipal HDI	2010 UNDP	2019 UNDP
	High level of education (gender and ethnicity)	2010 IBGE	2018 IBGE
	Average income (gender and ethnicity)	2010 IBGE	2018 IBGE
	Unemployment rate	2010 IBGE	2018 IBGE
	Aging rate	2010 IBGE	2018 IBGE
Environmental risk factors and chronic diseases	Obesity and sedentary lifestyle (prevalence)	2013 PNS	2019 PNS
	Alcohol and tobacco (prevalence)	2013 PNS	2019 PNS
	Arterial hypertension and diabetes (prevalence)	2013 PNS	2019 PNS
	Adequate sanitation—proportion of population served by sewage or septic tanks, garbage collection and water supply	2010 IBGE	2018 IBGE

(Continued)

TABLE 1 (Continued)

Dimensions	Variables	Period 1 (*t*)	Period 2 (*t* + 1)
Governance	Transparency Level	2015 MPF	2016 MPF

Source: CIH, Hospital Information Communication; CNES, National Register of Health Establishments; DATASUS, Department of Informatics of the Unified Health System; e-manager-AB, PHC system; HDI, Human Development Index; IBGE, The Brazilian Institute of Geography and Statistics; MDS, Social Development Ministry; MPF, Federal Public Ministry; PNI, National Immunization Program; PNS, National Health Survey; SIA, Ambulatory Information System; SIH, Hospital Information System; SIAB; SISAB, Primary Care Health Information System; SIM, Mortality Information System; SINAN, Notifiable Diseases Information System; SINASC, Information System on Live Births; SIOPS, Information System for the Public Budgets in Health; SISCOLO and SISMAMA, Cervical and breast cancer screening information systems; STN/MF, National Treasury Secretariat/ Ministry of Finance; UNDP, United Nations Development Program.

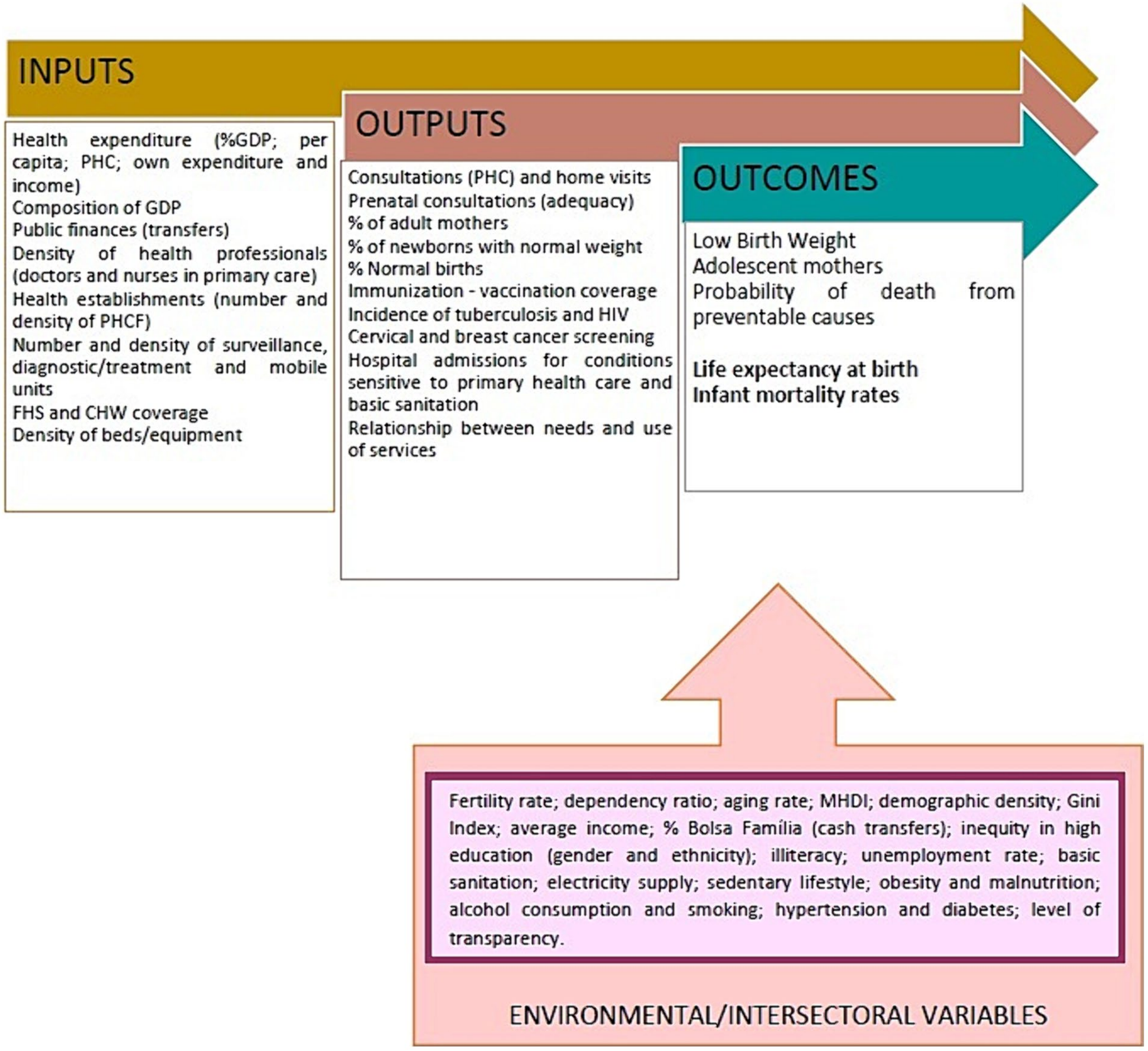


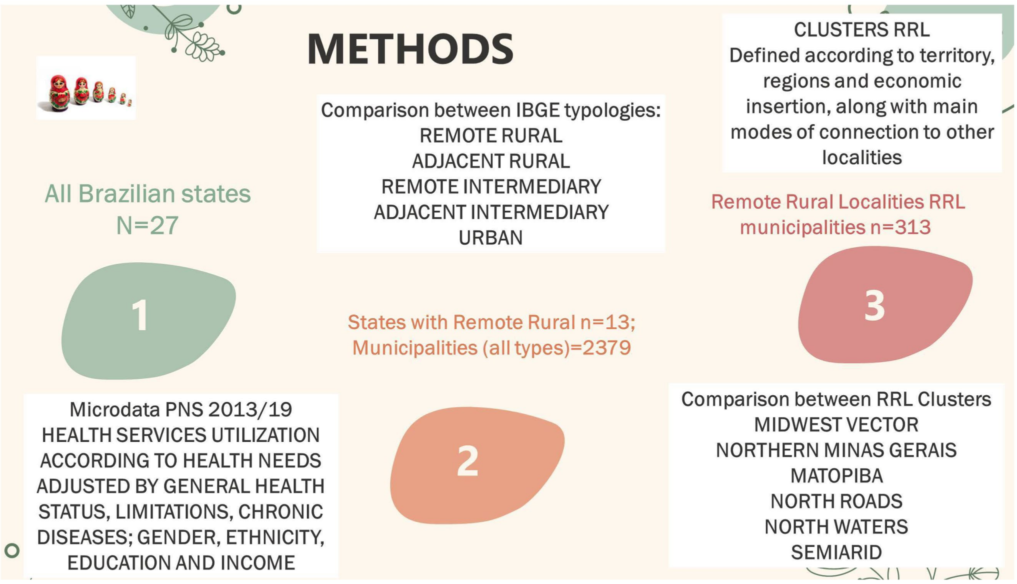
FIGURE 1
Health production model with the following variables: inputs or resources, products or intermediate results (outputs), final results (outcomes), and environmental results (throughput or control).

where $f(x) = \beta_0 + \beta_e X_e + \beta_{c1} X_{c1} + \beta_{ec1} X_e * X_{c1} \ldots + \beta_{cn} X_{cn}$

β_0 corresponds to the intercept; $\beta_e X_e$ are the equation terms, β is the coefficient and X refers to the independent exposure variables; $\beta_{c1} X_{c1}$ is the covariable 1 (until n, $\beta_{cn} X_{cn}$); $\beta_{ec1} X_e * X_{c1}$ is the interaction term between the exposure variables and covariable 1 (confounding variable).

The sample size is about 10 to 30 observations per variable. We applied the *stepwise backward* method, from the initial complete model, until managing to find the adequate final model. The analyses were carried out using *Stata SE 14.0* software.

We then proceeded to use the fixed effects regression model for panel data (time-invariant characteristics) including confounding and



METHODS

All Brazilian states
N=27

1

Microdata PNS 2013/19
HEALTH SERVICES UTILIZATION
ACCORDING TO HEALTH NEEDS
ADJUSTED BY GENERAL HEALTH
STATUS, LIMITATIONS, CHRONIC
DISEASES; GENDER, ETHNICITY,
EDUCATION AND INCOME

Comparison between IBGE typologies:
REMOTE RURAL
ADJACENT RURAL
REMOTE INTERMEDIARY
ADJACENT INTERMEDIARY
URBAN

States with Remote Rural n=13;
Municipalities (all types)=2379

2

CLUSTERS RRL
Defined according to territory,
regions and economic
insertion, along with main
modes of connection to other
localities

Remote Rural Localities RRL
municipalities n=313

3

Comparison between RRL Clusters
MIDWEST VECTOR
NORTHERN MINAS GERAIS
MATOPIBA
NORTH ROADS
NORTH WATERS
SEMIARID

FIGURE 2
The three steps in our methods.

interaction variables in the effectiveness analysis, which considers the regression's beta coefficients, whilst the location-specific effect was calculated by adding the fixed effect to the output residuals, which calculates efficiency. We employed this method to all three stages listed below. The analyses were carried out using *Stata SE 14.0* software.

The equation below was used to obtain the final models:

$$Y_{it} = \beta_0 + \beta_1 X_{(1,it)} + \cdots + \beta_k X_{(k,it)} + y_2 E_2 + \cdots + y_n E_n + \delta_2 T_2 + \delta_t T_t + \mu_{it}$$

where Y_{it} is the dependent variable, e.g., Life expectancy at birth and Infant Mortality rates (DV where i = unit and t = time); $X_{(k,it)}$ represents the independent variables (VI), β_k is the coefficient for the VIs, u_{it} i is the error term, ε_n is the unit n. γ_2 is the coefficient for the units. T_t is the time, and δ_t is the coefficient relative to time. We have employed this method alone to compare Brazilian states (with and without RRL) and the typologies within RRL only and to compare urban-rural typologies along with DEA (in the states with RRL).

The fixed effects model has a relatively good correlation to the DEA analysis. All the same, we employed DEA dynamic model with stages to deepen the analysis of the states with and without RRL, with different dependent variables, to ensure validity for the models. Besides life expectancy at birth and infant mortality rates, we also had models for the probability of deaths due to preventable causes, the proportion of newborns with low birth weight, and the proportion of adolescent mothers.

The equation for the network dynamic model is derived from the slack model, product-oriented with k steps:

$$\frac{1}{\tau_0^*} = \max \Sigma_{k=1}^{K} W^k \left[\left[1 + \frac{1}{r^k + \Sigma_{h \in F_k} t_{k,\,h}} \left(\frac{\Sigma_{kr=1}^{r} s_{r0}^{k+}}{y_{r0}^{k}} + \frac{\Sigma_{h \in F_k} s_{h0}^{(k,h)+}}{z_{h0}^{(k,h)}} \right) \right]\right]$$

where, w_k is the relative weight of each division; F_k is the set of stages with links (k, h); $\sum K_k = 1\ w_k = 1$; $w_k \geq 0$; s_k^+ are the output slack vectors; r_k is the number of outputs in stage k; $t_{(k,\,h)}$ is the number of products in the link between stage k and h; $s_{h0}^{(k,\,h)+}$ are the slack vectors of the links and z deals with the intermediate products. We performed these analyses with the *Max Dea 8 Ultra* (DEA software).

2.3 Data collection

All our data were secondary, aggregated and public databases, used in an ecological perspective, except for the use of health services according to health needs, which was extracted from the National Health Survey.

The National Health Survey was carried out by the Ministry of Health, together with the Brazilian Institute of Geography and Statistics. The survey was carried out with a probabilistic sample of households, with primary sampling units, composed by at least one census sector and 12–18 households, according to the number of households in every state. In each household, a resident (15 years or older) was randomly selected and interviewed on behalf of the group living together, with questions about lifestyle, work, chronic diseases and violence. The method employed at all stages was the simple random sample. Data were collected during 2019–2020 from 108,457 households.

2.4 The three analyses carried out

2.4.1 All Brazilian states

First, an analysis of efficiency and effectiveness was carried out with all Brazilian Federative Units (26 states and 1 Federal District), including resource, health, and intersectoral categories, using the fixed effects model. We adopted the health production model, with life expectancy at birth (LEB) and infant mortality rates (IM) as effect variables, in two stages. It is noteworthy that the models relating to the results of health services relate to the use of services according to health needs. We included microdata from the 2013 and 2019 National Health Survey-PNS in the database to circumvent the bias of using ecological variables (37). We compared the states with and without RRL as the first level of analysis.

We calculated the use of health services at the state level, according to health needs, by regressions in which we tested the use of services, general health status, and limitations resulting from chronic diseases, adjusting for gender, race, marital status, education, and income. The variables of age, chronic diseases (collinear), and enrollment in FHS units were not statistically significant.

2.4.2 Brazilian states with remote rural localities (municipality level)

Then, we carried out efficiency and effectiveness analyses using the EF and DEA models, comparing the remote rural typology with the others (2). We performed a controlled comparison among the states with RRL between the remote rural typology and the typologies of rural adjacent, intermediate adjacent and remote, and urban. For further details, please check Supplementary material S2.

This analysis considered only the Brazilian states with RRL ($n = 13$; 2,379 municipalities). The states with RRL comprise all seven states in the north region, three states in the northeast region (Bahia, Maranhão, and Piauí), and two in the Midwest region (Mato Grosso do Sul and Mato Grosso), in addition to Minas Gerais, the only one in the southeast region. Three were excluded, due to low representativeness of their units (2 in the South and 1 in the Northeast).

The dependent variables in these models were, in addition to life expectancy at birth and infant mortality, the probability of deaths due to preventable causes, the proportion of newborns with low birth weight, and the proportion of adolescent mothers. In this way, we decided to evaluate the intermediate health results (outputs), which could relate in a more detailed and proximal way to the different organizational arrangements present in these locations, and the results of efficiency and effectiveness in health outcomes.

2.4.3 Remote rural localities (municipalities) only

In addition, we also studied the typologies of the different RRL among themselves. This typology was elaborated by Bousquat et al. (4), starting from the category of analysis of the territory use and having as reference the study of Santos and Silveira (38) and the identification of four regions, the concentrated one (South and Southeast) the one of Recent Peripheral Occupation; the Northeast and the Amazon. Bousquat et al. (4) then classified the RRL by the respective logic of the economic circuit insertion and their prevalent form of interconnection with the other points of the territories, whether by land or river.

Next, Bousquat et al. (4) scanned the RRL and broke them down by variables that denote the rarefaction and remoteness of the population, in addition to economic capacity. The qualitative analysis led to the design of six clusters (313 RRL or 97% of the total) namely: Matopiba ($n = 92$); Northern Minas Gerais (NMG) ($n = 22$); Midwest Vector (MWV) ($n = 84$); Semiarid ($n = 42$); North Waters ($n = 45$); and North Roads ($n = 28$). For more information, please refer to Supplementary material S3.

Finally, we compared the different RRL clusters with each other (4), with regard to intersectionality. Thus, we compared the Midwest Vector, Matopiba, Northern Minas Gerais, North Roads, North Waters, and Semiarid concerning the reach of efficiency and effectiveness in health, with equity as a reference to these relationships.

We compared the typologies of the municipalities and the RRL clusters using the Kruskal–Wallis test and the multiple comparisons with the Dunn test, and Bonferroni adjustments.

We performed the analyses with *Stata SE 10.1 and 14.0* software (Fixed Effects model) and *Max Dea 8 Ultra* (DEA).

3 Results

We present the results in three levels of analysis, which allowed us to deepen our understanding of the RRL and their health needs relating to their territories and contiguities. The main summary results are available in Figure 3. Next, we give more details on each analysis result.

3.1 Comparison between Brazilian states with and without RRL, considering health needs

First, we performed bivariate analyses on the dimensions according to the fixed effects model. We retrieved data on service utilization from the 2013 and 2019 National Health Surveys (PNS) (37) based on the relationship between health needs and the use of services (6, 7). We selected the fixed effects model because of its ability to capture health effectiveness through its coefficients and efficiency through the analysis of residuals. The data on the use of services was essential to reduce the gap between needs and the utilization of services, with less aggregation of health data, which usually presents very low determination coefficients.

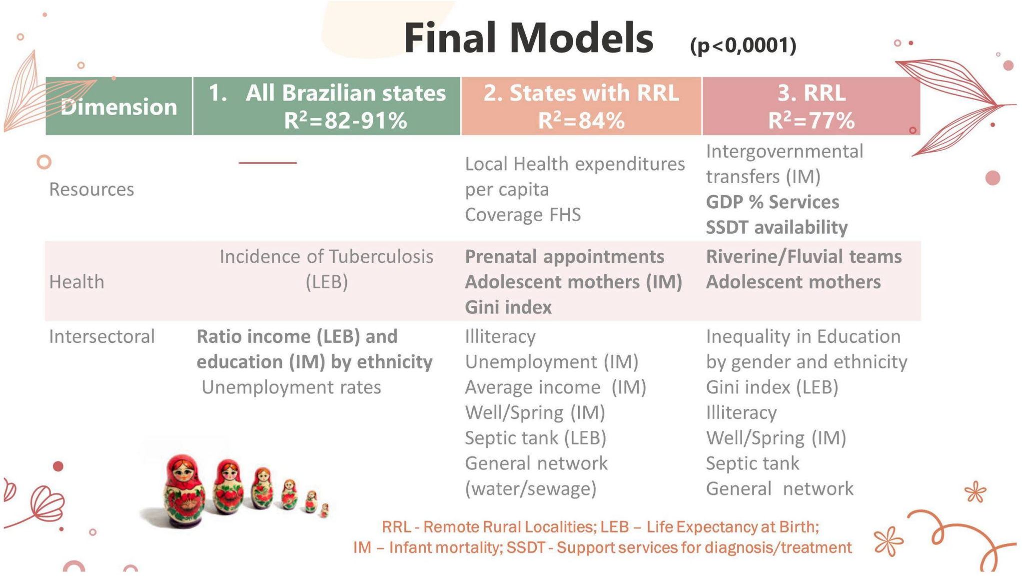
The highest use of services occurred among those with limited functional capacity to carry out activities of daily living, with worse general health status, female, white, married, and with a better level of education and higher income (Table 2). The variables age, chronic diseases, and registration with the FHS did not remain in the final model.

Based on the bivariate analysis (Supplementary material S4), we selected the variables that would be part of the multivariate analysis according to their statistical significance and coefficient value concerning other variables in their dimension and group.

The results of the comparison of Brazilian states with and without the RRL (Table 3) revealed high determination coefficients ($R^2 = 82–96\%$), drawing attention, especially to the health dimension, which had high values both in the efficiency and productivity analysis, i.e., the relationship between resources and services (61–93%), very similar to the other dimensions (Table 4). These results were even more significant for service utilization, ranging from 77–82%. Moreover, in the final model for life expectancy at birth, the variable incidence of tuberculosis remained a relevant variable.

Most strikingly, the final models for both effects retained variables relating to ethnic inequity in income (LEB) or education (IM) in conjunction with unemployment rates. Thus, the final models are more intersectoral, with no resource dimension variables remaining.

Regarding life expectancy at birth and infant mortality, the best results are in the southeast, part of the south and the capital, besides a small part of the Amazon region (LEB only). Considering the dimensions, the resources and health dimensions present better results for the North and Northeast regions, whereas the intersectoral



Final Models (p<0,0001)

Dimension	1. All Brazilian states R^2=82-91%	2. States with RRL R^2=84%	3. RRL R^2=77%
Resources	—	Local Health expenditures per capita Coverage FHS	Intergovernmental transfers (IM) **GDP % Services** **SSDT availability**
Health	Incidence of Tuberculosis (LEB)	**Prenatal appointments** **Adolescent mothers (IM)** **Gini index**	**Riverine/Fluvial teams** **Adolescent mothers**
Intersectoral	**Ratio income (LEB) and education (IM) by ethnicity** Unemployment rates	Illiteracy Unemployment (IM) Average income (IM) Well/Spring (IM) Septic tank (LEB) General network (water/sewage)	Inequality in Education by gender and ethnicity Gini index (LEB) Illiteracy Well/Spring (IM) Septic tank General network

RRL - Remote Rural Localities; LEB – Life Expectancy at Birth;
IM – Infant mortality; SSDT - Support services for diagnosis/treatment

FIGURE 3
Summary results of the three analyses for life expectancy at birth and infant mortality.

TABLE 2 Regression of use of health services according to selected sociodemographic and health variables.

Variable	OR (IC 95%)	p
Limited functional capacity (LFC)—ref (yes)	0.63 (0.59–0.66)	<0.001
General health status ref (better)	1.46 (1.41–1.51)	<0.001
Sex (ref: male)	1.42 (1.35–1.48)	<0.001
Race (ref: white)	0.98 (0.96–0.99)	0.019
Marital status (ref: married)	0.96 (0.94–0.98)	<0.0001
Education level (ref: lower)	1.07 (1.05–1.09)	<0.001
Income level (ref: lower)	1.11 (1.09–1.14)	<0.001
Constant	0.12 (0.10–0.15)	<0.001

$N = 77.887$; Population 85,801,346; $F = 207.12$; $p < 0.0001$.

dimension shows higher scores for the southeastern region and the capital, besides a small part of the Amazon region (LEB). To find out more details, see Supplementary material S5.

It is worth mentioning that the level of transparency [Federal Public Ministry (MPF)] was highly significant in the bivariate analysis in the expected direction, although it did not remain in the dimensional models. In contrast, income inequality by gender was statistically significant in the opposite direction and did not remain in the final models.

We also present graphs showing the potential gains in years of life or the reduction in infant mortality rates attributed to efficiency gains by state and macro-region for the set and the dimensions analyzed (Supplementary material S5).

The comparison between the 13 states with and without RRL ($n = 14$) on Graphs 1A,B shows that at the level of resources and health, there is greater efficiency in states with remote rural municipalities ($p \leq 0.01$), while regarding the intersectoral and global models, the reverse is perceived, being significant for infant mortality ($p \leq 0.01$).

We also performed a technical efficiency analysis (productivity), associating the physical and financial resource variables with the health dimension variables, as well as the intersectoral variables. The coefficients of determination were very high (Table 4), ranging from 61–93% for the resource dimension and 59–90% for the intersectoral dimension. The lowest values were for the tuberculosis incidence variable, whereas the highest was for the proportion of vaginal births. The most relevant variables were health spending as a % of GDP, the % of the population earning less than ¼ of the minimum wage, and the % of GDP allocated to Public Administration. Concerning the intersectoral variables, the Gini index, ethnicity inequality of higher education, the unemployment rate, and inadequate sanitation stand out.

Regarding the health variables (Table 4), we depict that the use of health services was negatively associated with low income and positively associated with the proportion of GDP of the Public Administration and employment rates. Thereby, the states containing RRL presented lower utilization than the others. It is worth mentioning that the incidence of tuberculosis, hospitalizations for sanitation-sensitive causes, vaginal birth deliveries, and smoking were influenced by ethnicity inequalities in education and the proportion of the population with low income, according to variables remaining in the dimensional models. Furthermore, health expenditure as a % of state GDP was relevant for the incidence of tuberculosis (along with unemployment and lower proportions of GDP derived from the Public Administration) and for hospitalizations due to sanitation-sensitive causes. Income inequalities (Gini index) were relevant for the higher proportion of vaginal birth deliveries and smoking prevalence.

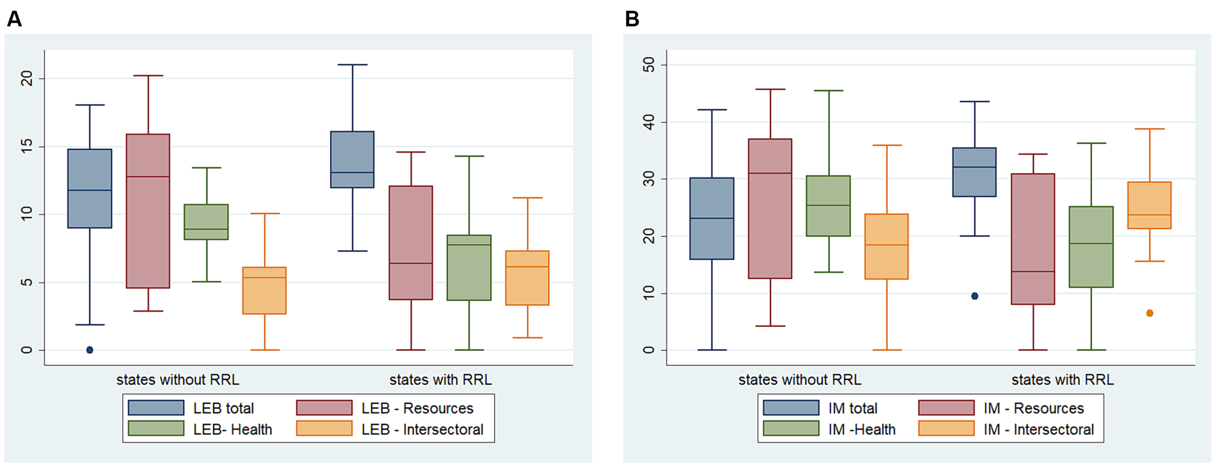
TABLE 3 **Final regression models for selected effect variables, according to dimensions and general models (model with all states: Brazil, 2010 and 2019).**

Model/variable	Life expectancy at birth—LEB variable (beta)	Infant mortality—IM variable (beta)
Physical and financial resources	% population receiving < ¼ MW (−0.40) Health expenditure % GDP (−1.33) Public administration % GDP (6.74×10^{-5}) Constant (87.00) ($R^2 = 96\%$; $p < 0.0001$)	% population receiving < ¼ MW (1.38) Density of nurses (−3.24) Public administration % GDP (-1.55×10^{-7}) Constant = −14.83 ($R^2 = 96\%$; $p < 0.0001$)
Health production	Use of health services according to health needs (0.41) TB incidence (−0.10) Smoking (−0.36) General proportion of normal deliveries (−0.29) Constant (88.14) ($R^2 = 95\%$; $p < 0.0001$)	Use of health services according to health needs (−1.94) Hospitalizations sensitive to basic sanitation (2.49) Smoking (2.68) Constant (8.34) ($R^2 = 86\%$; $p < 0.0001$)
Intersectoriality	Ratio ethnicity income (−5.60) Ratio ethnicity education (−1.93) Unemployment rate (−0.40) Inadequate sanitation (−0.22) Constant (94.44) ($R^2 = 92\%$; $p < 0.0001$)	Gini index (122.20) Ratio ethnicity education (11.42) Unemployment rate (0.96) Constant (−93.97) ($R^2 = 85\%$; $p < 0.0001$)
General—all dimensions	Ratio income ethnicity (−7.91) Unemployment rate (−0.44) TB incidence (−0.15) Constant (96.96) ($R^2 = 91\%$; $p < 0.0001$)	Ratio ethnicity education (12.14) Unemployment rate (1.53) Constant (26.94) ($R^2 = 82\%$; $p < 0.0001$)

TABLE 4 **Final regression models for health variables, according to the dimensions of resources and environmental variables (model with all states: Brazil, 2010 and 2019).**

Model/variable	Physical/financial resources variable (beta)	Intersectoriality variable (beta)
Service utilization	% population that receives < ¼ MW (−0.14) Public administration % GDP (6.23×10^{-8}) Constant (18.10) ($R^2 = 77\%$; $p < 0.0001$)	Unemployment rate (−0.46) Constant (20.16) ($R^2 = 82\%$; $p < 0.0001$)
Incidence tuberculosis TB	% population that receives < ¼ MW (−0.14) Health expenditure % GDP (6.46) Public administration % GDP (-1.91×10^{-7}) Constant (−6.83) ($R^2 = 61\%$; $p < 0.0001$)	Ratio ethnicity education (7.01) Unemployment rate (0.99) Constant (9.27) ($R^2 = 59\%$; $p < 0.0001$)
Hospitalizations BSSC*	% population receiving < ¼ MW (0.17) Health expenditure % GDP (1.03) Constant (−5.44) ($R^2 = 81\%$; $p < 0.0001$)	Ratio ethnicity education (1.22) Inadequate sanitation (0.20) Constant (−2.10) ($R^2 = 70\%$; $p < 0.0001$)
Normal deliveries (%)	% population receiving < ¼ MW (0.65) Constant (32.60) ($R^2 = 93\%$; $p < 0.0001$)	Gini index (124.62) Ratio ethnicity education (4.51) Inadequate sanitation (0.31) Constant (−42.00) ($R^2 = 90\%$; $p < 0.0001$)
Smoking (%)	% population receiving < ¼ MW (0.17) Constant (8.41) ($R^2 = 72\%$; $p < 0.0001$)	Gini index (32.06) Ratio ethnicity education (1.73) Constant (−11.21) ($R^2 = 63\%$; $p < 0.0001$)

*MW, minimum wage; BSSC, basic sanitation-sensitive causes.



GRAPH 1
(A) Distribution of potential years of life gained attributed to efficiency gains according to states with and without remote rural municipalities.
(B) Distribution of the potential reduction in infant mortality due to improved efficiency according to states with and without remote rural municipalities.

3.2 Comparison of efficiency among the states with RRL, between the remote rural typology and the urban, adjacent or remote intermediate, and adjacent rural

In the second analysis, which involves only the states with RRL, we combined the fixed effects model with network and dynamic DEA. This process yielded satisfactory correlations of the efficiency scores from both models, higher for the indicators closest to health services (intermediate results), such as the deaths from preventable causes, the % of low birth weight, and the % of teenage mothers. Once again, we used bivariate analyses to select the variables considered for the multivariate models by dimension and group of variables (Supplementary material S6).

The models regarding health levels (LEB and IM) with only the 13 states containing RRL remained with the Gini index, local health expenditure *per capita*, and FHS coverage, in addition to basic sanitation (general network, septic tank, and well/spring), illiteracy and unemployment (IM). Additionally, the variables related to adequate prenatal care and the proportion of teenage mothers (IM) stand out. Remarkably, the determination coefficients were 84%, presenting a regular correlation between the methods of FE and DEA of 46% (Tables 5, 6).

The family health and oral health teams and the CHW showed relevant effectiveness results in reducing *preventable deaths*, combined with employment, adequate prenatal care, adequate birth weight, and treatment for acute respiratory infections and diarrhea in children under 2 years of age.

Regarding *low birth weight*, the following variables remained relevant: home visits by the Family Health Team, *per capita* health expenditure, and the proportion of primary care physicians.

Concerning the proportion of *teenage mothers*, intergovernmental transfers are a fundamental resource variable, as well as the density of primary care physicians, high education, average income, and income inequality (Gini index).

The determination coefficients were much lower than in the health levels models precisely because they represent intermediate output variables (2–38%). On the other hand, they showed a high correlation between the methods employed (FE and DEA), ranging from 87–97% (Tables 5, 6).

It is possible to see the distribution of these results (Supplementary material S7) for LEB, IM, deaths from preventable causes, % of low birth weight, and adolescent mothers, by typology and by state, with the potential gains in years of life or the reduction in the other measures (rates and proportions) due to improved efficiency.

The RRL stand out compared to the other typologies, especially regarding avoidable deaths, LEB, and IM (Table 5). On another note, the RRL exhibited a tie with the remote intermediate municipalities and a second-place position after the adjacent rural municipalities for low birth weight and the proportion of adolescent mothers. We may then show a notable gradient among typologies for the set of models: the RRL with the best values, followed by the adjacent rural, remote intermediate, adjacent intermediate, and, lastly, urban. To check the Brazilian states ranking, please refer to Supplementary material S7.

Remote localities (intermediate and rural) have higher distributions of overall income inequality (Gini index) and income inequality by gender relative to adjacent municipalities (intermediate and rural). On the other hand, the differences in education levels by gender and ethnicity are lower in remote rural municipalities than in the remaining typologies and represent a determining factor for their efficiency and effectiveness.

3.3 Comparison between remote rural clusters regarding efficiency and effectiveness, with exclusive models intersectionality-wise

In the third analysis, we evaluated the RRL clusters only with the multivariate fixed effects analysis since we had already assessed the regression models and knew which relevant variables to test the final models.

It is interesting to compare the different models, as if in layers, according to the weight of the RRL (Table 7). When we analyze all the Brazilian states, we realize that the final model encompasses

TABLE 5 Position in relation to the average rank of efficiency for each geographic classification (model with 13 states containing remote rural localities).

Geographic classification*	LEB	IM	Preventable deaths (%)	Low birth-weight (%)	Teen mothers (%)	Average rank
Remote rural (RR)	2.2 (1)	2.0 (1)	1.3 (1)	2.8 (1.5)	2.6 (2)	2.2 (1)
Urban (U)	4.6 (5)	4.8 (5)	4.2 (5)	3.2 (4.5)	3.3 (3)	4.0 (5)
Adjacent rural (AR)	2.7 (3)	2.5 (2)	2.8 (2)	3.2 (4.5)	2.0 (1)	2.6 (2)
Remote intermediate (RI)	2.6 (2)	2.8 (3)	3.1 (3)	2.8 (1.5)	3.7 (5)	3.0 (3)
Adjacent intermediate (AI)	2.8 (4)	3.0 (4)	3.7 (4)	3.1 (3)	3.5 (4)	3.2 (4)
Remaining variables in the final model	$n = 7$	$n = 9$	$n = 6$	$n = 3$	$n = 5$	$n = 19$
FHS coverage	×					1
FHS % teams			×			1
CHW			×			1
FHS home visits				×		1
% PHC physicians				×	×	2
Gini	×	×			×	3
% Illiteracy	×	×				2
% High education					×	1
% Unemployment		×	×			2
Average income		×			×	2
Diarrhea/ARI < 2y			×			1
Prenatal consultations	×	×	×			3
Low birthweight			×			1
% Adolescent mothers		×				1
Septic tank	×					1
Well/spring		×				1
General network	×	×				2
Local health expenses	×	×		×		3
Intragov transfers					×	1
R^2	84%	84%	10%	2%	38%	$p < 0.0001$
Correlation FE and DEA	47%	46%	96%	97%	87%	$p < 0.0001$

Multiple comparisons—Dunn test $p = 0.0001$; LEB AI/AR > RI/RR; U > all; IM AI > RI/AR/RR; AR > RI/RR. U > other; preventable deaths AI > RI/AR > RR. U > all; low birthweight AI > RI/RR; AR > RR. U > all; adolescent mothers IR > all; RR > AI > AR; U > AR; RR > U. ARI, acute respiratory infection; CHW, Community Health Workers; EPC, local health expenses per capita; FHS, Family Health Strategy; IM, infant mortality; PN, prenatal; LEB, life expectancy at birth.

intersectional and health issues concerning the incidence of tuberculosis. When we repeat the model with only the 13 states containing RRL, we verify emerging variables regarding income inequality (Gini index), resources (health expenditure *per capita*) and FHS coverage, basic sanitation specific to these localities (general network, septic tank, and well/spring), illiteracy and unemployment (IM). The variables related to adequate prenatal care and the proportion of teenage mothers (IM) draw attention.

When we analyze the RRL only (Table 7), the models denote the importance of riverine and fluvial health teams, the proportion of diagnostic and treatment units, very relevant in these localities, in addition to the proportion of illiteracy and teenage mothers, even in the LEB model. This situation, of the level of education, also manifests itself in the inequity of reaching high levels of schooling among gender and ethnicity (higher inequities observed in the clusters of the Midwest Vector and North Waters). The proportion of the GDP derived from services is another relevant variable, which marks the living standards since they constitute advances allied to the agricultural activity in these localities. All models showed high determination coefficients (77–81%), especially in the models with more states.

The best efficiency results (LEB) were observed in the Amazon region, in the North Roads and Waters clusters, in that order, with relatively lower values of potential years of life gained by improved efficiency compared to the other typologies. Regarding the IM efficiency model, North Roads stands out along with the Northern Minas Gerais cluster, reaching a distribution close to North Waters. The others (MWV, MATOPIBA, and Semiarid) present higher potential reductions in infant mortality rates (Graphs 2A,B).

The best results are in Acre, Pará, Amazonas, and Amapá, which are part of the North Roads and North Waters Amazon clusters for

TABLE 6 **Final regression models for selected effect variables, according to dimensions and general models (model with remote rural localities states only: Brazil, 2010 and 2019).**

Effect variable	Independent variables (beta)	Details
Life expectancy at birth	Local health expenses *per capita* (3.9×10^{-3})	Constant (67.48)
	Gini index (−4.24)	($n = 4.678$; $R^2 = 84\%$; $p < 0.0001$)
	FHS coverage (0.52)	
	Illiteracy (−0.25)	
	Prenatal appointments (5.36)	
	Septic tank (0.01)	
	General network (0.05)	
Infant mortality	Local health expenses *per capita* (−0.01)	Constant (38.58)
	Gini index (−12.36)	($n = 4.724$; $R^2 = 84\%$; $p < 0.0001$)
	FHS coverage (−1.69)	
	Illiteracy (0.67)	
	Prenatal appointments (−16.06)	
	Adolescent mothers (5.28)	
	Unemployment rates (0.08)	
	Average income (−0.02)	
	Well/spring (−0.07)	
	General Network (−0.16)	
Preventable deaths	Proportion of FHS teams and oral health (−0.40)	Constant (2.28)
	Proportion of CHW teams (−1.00)	($n = 3.379$; $R^2 = 10\%$; $p < 0.0001$)
	Prenatal appointments (0.73)	
	ARI/diarrhea <2a (−0.52)	
	Low birthweight (2.09)	
	Unemployment (−0.01)	
Low birthweight	Local health expenses *per capita* (7.35×10^{-6})	Constant (0.10)
	Domiciliary visits (−0.01)	($n = 3.800$; $R^2 = 2\%$; $p < 0.0001$)
	Proportion of medical doctors PHC (−0.02)	
Adolescent mothers	Intergovernmental transfers (-5.18×10^{-4})	Constant (0.32)
	Higher education (-2.08×10^{-3})	($n = 4.605$; $R^2 = 38\%$; $p < 0.0001$)
	Gini index (0.07)	
	Average income (-7.29×10^{-5})	
	Density of medical doctors PHC (−12.78)	

both effect variables, with a reduced number of potential years of life gained and lower potential reductions in the infant mortality rate due to efficiency gains. The state of Rondônia, in the MWV, also stands out (Graphs 2C,D).

The inequalities in low income (% of the population receiving less than ¼ of a minimum wage) were higher for the MWV cluster (higher than the other typologies) and Matopiba, compared to the Semiarid. The inequalities in education by gender and ethnicity, on the other hand, were higher for the MWV and North Waters clusters compared to the remaining typologies, except for the North Roads region. Concerning income inequality by gender, the MWV presented the worst results, and the Northern Minas Gerais the best.

We may summarize our most important results below, depicting in which settings the RRL score higher than their counterparts:

Lastly, you may have a full view of all the results and the variables used in Supplementary material S8, including metadata, which can help the interested reader understand and reproduce this analysis. We also offer a summary table of all the efficiency analyses carried out and the variables remaining in the final models.

4 Discussion

The contrasts among Brazilian states bring difficulties inherent to their level of aggregation. At this level, the summary measures are difficult to interpret but also have some advantages, especially regarding the existence of more robust measurements in health when arising from epidemiological surveys, when compared to the local and

TABLE 7 Comparison between the different models, according to data aggregation.

Model/Variable	Life expectancy at birth variable (beta)	Infant mortality variable (beta)
All Brazilian states	Income ethnicity ratio (−7.91) Unemployment rate (−0.44) Tuberculosis incidence (−0.15) Constant (96.96) ($n = 54$; $R^2 = 91\%$; $p < 0.0001$)	Education ethnicity ratio (12.14) Unemployment rate (1.53) Constant (−26.94) ($n = 54$; $R^2 = 82\%$; $p < 0.0001$)
RRL states only	Local health expenses *per capita* (3.9×10^{-3}) Gini index (−4.24) FHS Coverage (0.52) Illiteracy (−0.25) Prenatal appointments (5.36) Septic tank (0.01) General Network (0.05) Constant (67.48) ($n = 4.678$; $R^2 = 84\%$; $p < 0.0001$)	Local health expenses *per capita* (−0.01) Gini index (−12.36) FHS coverage (−1.69) Illiteracy (0.67) Prenatal appointments (−16.06) Adolescent Mothers (5.28) Unemployment rates (0.08) Average income (−0.02) Well/spring (−0.07) General Network (−0.16) Constant (38.58) ($n = 4.724$; $R^2 = 84\%$; $p < 0.0001$)
RRL only	Gini index (−4.39) GDP %-services (15.35) Riverine/fluvial teams (5.23) Inequality in education by gender/ethnicity (−1.06) Adolescent mothers (−9.73) Illiteracy (−0.29) Septic tank (0.03) General network (0.11) SSDT availability (4.69) Constant (72.78) ($n = 610$; $R^2 = 77\%$; $p < 0.0001$)	Intragovernmental transfers (−0.13) GDP % services (−48.07) Riverine/fluvial teams (−34.83) Inequality in education by gender/ethnicity (3.16) Adolescent mothers (20.74) Illiteracy (0.74) Well/spring (−0.13) Septic tank (−0.09) General network (−0.31) SSDT availability (−16.94) Constant (46.42) ($n = 608$; $R^2 = 77\%$; $p < 0.0001$)

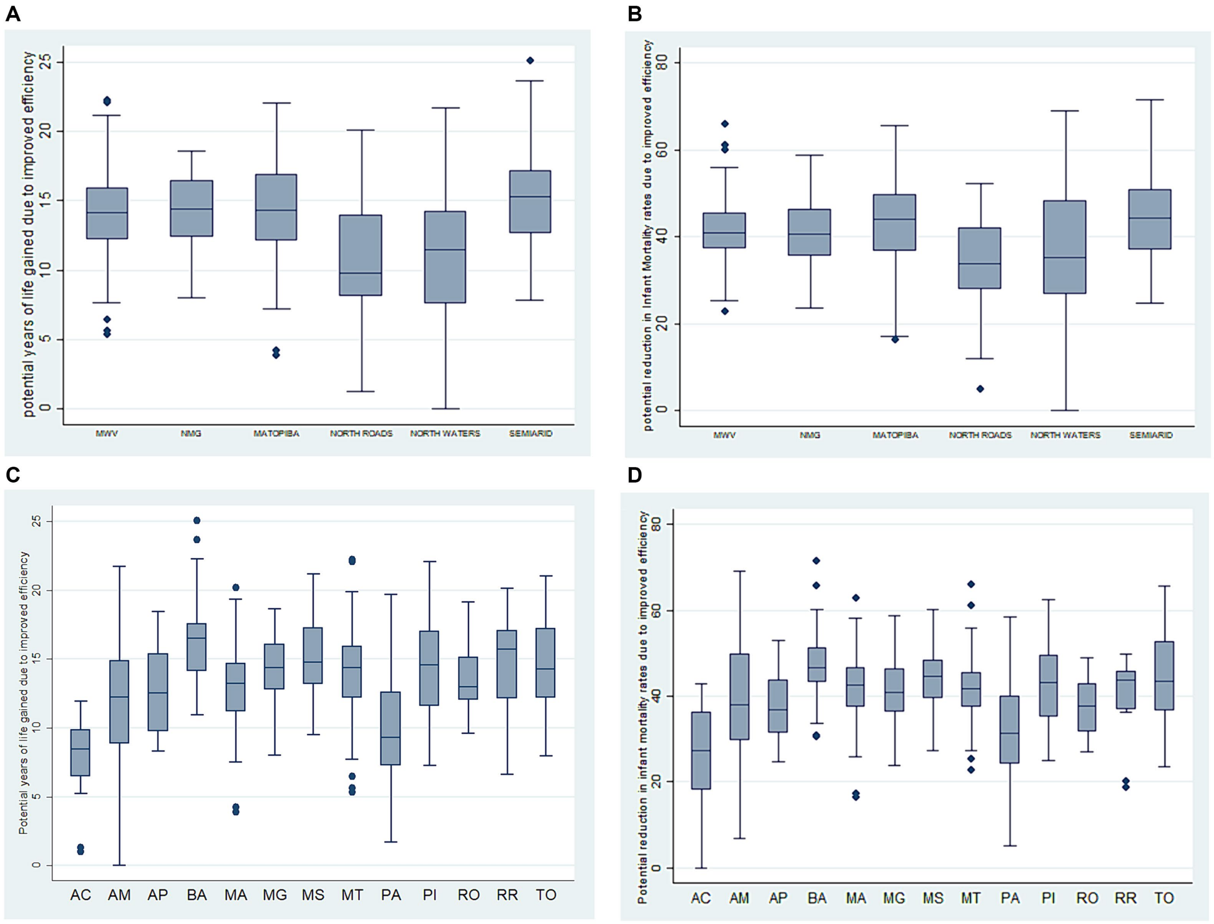
Brazilian states, states with remote rural localities (RRL and only Brazil, 2010 and 2019). *FHS, Family Health Strategy; SSDT, Support services for diagnosis and treatment.

RLL score better at resources and health levels Level of resources and health were more efficient and effective in RRL states, scoring less at the intersectoral/global level	**RRL, compared to the other rural-urban typologies scored better overall**, with PHC models, basic sanitation, local health expenditures, income equality, employment, literacy, adequate prenatal care and adult mothers, and less education inequalities by gender/ethnicity
RRL used less health services, according to health needs, which was associated with low income and lower level of GDP (Public Administration)	**The best organizational models** were the FHS with CHW working in the territory, with home visits, prevention of acute respiratory infections and diarrhea and primary care physicians, with adequate birthweight
Less inequities in RRL Incidence of tuberculosis, hospitalizations due to sanitation-sensitive health causes, vaginal birth deliveries and smoking were influenced by ethnicity inequities in education and % of population with low income, average income inequities and % state GDP	**Best results in the Amazon region** Riverine and fluvial health teams, with diagnostic and therapeutic services, with less disparity in education levels, less teenage mothers, high % of Services GDP

global levels (32). Thus, our study assessed efficiency and effectiveness in health, with the possibility of including the utilization of health services according to health needs, as microdata from the National Health Survey.

Unlike the analyses performed at the global and local levels, we demonstrated the potential to dimension health more accurately. The differences observed shed light on the issue that healthcare is more closely related to health outcomes when related to health needs. In other words, by being disconnected from health needs, health actions do not contribute much to the health levels of society (6).

It is important to ponder that the results in the southeastern region do not mean that the states in this region are in a better situation than the others. We may deduce that in these places, the expected results are worse than the observed ones, such as the high levels of inequality, the incidence of tuberculosis, and unemployment. Thus, they could be much worse off than they are. This statement leads us to another



GRAPH 2
(A) Potential years of life gained, due to increased efficiency in the clusters of remote rural municipalities in Brazil. **(B)** Reduction in infant mortality rates due to efficiency gains in the clusters of remote rural municipalities in Brazil. **(C)** Potential years of life gained attributed to increased efficiency in Federative Units with remote rural municipalities in Brazil. **(D)** Reduction in infant mortality rates due to efficiency gains in the Federative Units of remote rural municipalities in Brazil.

reflection: are these places increasing their average results at the expense of gender and ethnic inequalities in income and education? Considering the contemporary capitalist development model, these inquiries make much more sense than we would like to admit (39).

The differences observed among the RRL states evidenced, in turn, greater efficiency in the dimensions of resources and especially health, with more promising results than intermediate and urban localities. Therefore, the models containing health expenditures, the % of GDP of the Public Administration, the density of nurses, and the proportion of low-income achieved better results in these localities. Concerning health, likewise, the final model showed better results in the states with RRL through the utilization of services according to health needs, the incidence of tuberculosis, the proportion of vaginal deliveries, smoking, and sanitation-sensitive hospitalizations.

We observed that reducing inequalities in income and education by ethnicity could confer efficiency and effectiveness gains in the societal health levels. Reducing the incidence of tuberculosis, which involves a series of socioeconomic structural measures, and increasing the possibilities of formal employment and guaranteed labor rights are fundamental steps on this path (40). We must consider that these inequities are exceedingly high in some Brazilian states, reaching more than twice in the case of income and almost four times in education. This unfair distribution affects all of society, involving those in a position of apparent privilege.

It is interesting to observe some continuity between remote rural, adjacent rural, and remote intermediate as if rurality and remoteness maintain some overlapping in remote rural municipalities. The most striking differences between rural and remote recently discussed by Wakerman et al. (3) were the geographical aspect, isolation, access to general and health services, smaller populations and of original peoples, differentiated care models with smaller, more locally integrated teams, and a broader scope of actions with difficulties of specialist support, relevant socioeconomic challenges, reduced power and more difficulty in obtaining the necessary resources.

According to research on intersectionality in health (41), “place” does not carry a unique meaning or singular experience for participants. Three themes related to place were the most prominent in their data set: (i) location, (ii) distance/proximity, and (iii) urban or rural situation. Place refers to the actual locations where care takes place, while distance/proximity refers to the relational aspect of place, for example, in terms of being near or far from health services.

Situation refers to a sense of socially "positioned" or "situated" place. Intersectionality theory reminds us that these categorical findings are highly fluid and relational, and they complexly interrelate at various scales ranging from the body to overarching economic and sociopolitical structures (27).

Understanding place through an intersectional lens can increase the sophistication of the concept by raising questions about how researchers situate themselves in their research and develop, categorize, and understand the relationships between various types of places (28).

Intersectionality also increases the complexity of how identities are understood, made, undone, and simultaneously experienced in particular places. Similarly, a more sophisticated application of the concept of place to intersectional research can increase intersectionality's appreciation of social constructions and meanings of place and its role in shaping processes of oppression and subject formation as it shapes places (28).

Furthermore, social constructions of meaning and power infuse places and integrate a complex web of intersecting categories of difference (e.g., cultural, economic, historical, and political). Hence, these intersections will ultimately shape places, social experiences, and contexts of social interactions. We must understand gender roles relationally to place, with material and symbolic consequences on rural women's health. Gender and caregiving relations ultimately resonate with idealized views of the rural as an idyllic and feminine space, reinforcing and reproducing spaces of power and oppression (30, 42).

The remaining variables in the final RRL models point to the local reality for socio-environmental variables and those specific to healthcare models, such as basic sanitation with wells, springs, and septic. In these places, the general network is not always present and thorough, and the water supply and sewage collection occur in alternative ways, especially in the more distant regions. The health teams, the FHS, the CHW, and the fluvial and riverine teams stand out, as well as the importance of education and adequate prenatal care.

The remarkable theoretical framework of Bourke et al. (1), anchored in Giddens' social structuring, already emphasized the importance of local health responses from health professionals, managers, and users in the structuring of the local health system, buoyed by health systems in a broader sense, i.e., the social practices and the set of protocols and norms that homogenize the sociocultural space.

Another relevant point is interculturality regarding teenage pregnancy in these localities, especially the more remote and rarefied, which acquires importance in the observed models (43, 44). Proper prenatal care becomes even more relevant—when we consider adolescent pregnancy, which is more prevalent in rural areas and vulnerable populations—to ensure maternal-fetal well-being during the perinatal period and afterward. The undesirable consequences, such as low birth weight, remain throughout these children's lives.

The proportion of the services' GDP reinforces the different populations and activities in rural and remote localities, which present different economic and sociocultural insertions and which have been advancing in the services sector, including technological innovations with agricultural, extractive, fishing, and handicraft activities. They may belong to traditional communities such as indigenous, "quilombolas" (maroon), and river dwellers who reside in rural settlements or work at the headquarters as salaried employees (5).

Strikingly, the Amazon clusters (North Roads/Waters) showed the best efficiency results, although they have immense difficulties with geographical accessibility and less favorable health indicators (45). However, considering their local realities, they always seek to deliver health care to their populations, no matter how dispersed they are, besides expanding their clinical performance, acquiring critical inputs, and performing exams in their territory when there is a pressing need.

Whereas North Roads presents itself as a new agribusiness frontier region with a more structured regional health network, North Waters has no interconnection to the intermediate urban network in part of the RRL and connects directly to the metropolis, Manaus (46). Both clusters present rarified populations, a higher proportion of traditional peoples, with distinct ways of life and socioeconomic insertions (4, 5).

The organization of life in Amazonas features the centrality of Manaus, around which the other municipalities disperse. The capital monopolizes life and the provision of services with low interaction among the municipalities, causing the remote population to reach Manaus for healthcare purposes. The model of inter-municipal relations is asymmetric, vertical, and fragile (46).

Our results show that these two clusters use their resources efficiently, combining care management strategies adapted to their socioeconomic, demographic, and cultural reality, even by necessity, in the face of the asymmetric model of municipal relations to all types of services.

Concerning infant mortality, the Northern Minas Gerais region also presented more favorable efficiency and effectiveness results than the other clusters. This performance is due in part to the results already achieved in this locality, despite its socioeconomic vulnerability (12), with better maternal and child health indicators, as well as the high coverage of the FHS in its territory (4, 5) and, mainly, by the higher equity of income and education by gender and ethnicity.

Regarding organizational models, it is evident that coverage by FHS teams and CHW determine the efficiency and effectiveness of health actions when comparing remote rural municipalities to others in the same Federative Unit. In this sense, Castro et al. (47) demonstrated, in a case study, greater cost-effectiveness in the FHS than in traditional PHC facilities.

However, when we evaluate the RRL only, the fluvial and riverine teams become more important and visible, as the models demonstrate. This result highlights the importance, in the RRL, of the teams moving to where the population lives, often surrounded by water and in areas of elevated population rarefaction, respecting interculturality.

The local availability of biochemical and imaging tests and therapeutic support are essential factors in the coordination of care by PHC and the perception of healthcare satisfaction by users (48). Their offer can make a big difference in these regions since the time spent, the excessive traveling needs and high cost of performing exams in another municipality, and or the acquisition of medicines in distant public or private facilities can affect the health status and the results achieved by health services.

The Amazon territories engender new reflections on the best choice of the organization model to face the local realities and the users' needs. We should consider that the implementation of fluvial and riverine teams, as well as fluvial PHC facilities, mark relevant innovations in the way of organizing care management but introduces new needs for modifications in work processes and territorialization, with a family focus, markedly intercultural and anchored in health surveillance (10).

The investments required for its implementation may be higher than usual; however, the running costs are lower with more concrete results once the service adapts to the population and the use of the territory. Thus, these experiences can re-signify the best way to act in the territory: should the population be ascribed to the service, or should the service serve the territory? It reinforces the need for a strong, accessible, responsive, and proper (effective) PHC, offering continuity of care (49) and a community-territorialized approach that meets local needs (50).

In a case study in the Amazon region, Ferreira (51) defined the proportion of costs of a fluvial PHC facility, showing that most of them relate to human resources (64.62%), followed by inputs for health actions (17.72%), fuel (12.11%), and others (5.54%). We should draw attention to the potential efficiency gain from the performance of laboratory tests, at least the simplest ones, and from the displacement costs already planned and allocated in the scope of this kind of facility.

However, these arrangements require more planning and logistics to reach the fluid populations due to the activities and the water cycle with floods and ebb tides, characterizing challenges to overcome the distances and differentiated forms of displacement. Hence, the population's lifestyles already incorporate these fluctuations, which may facilitate or hinder access to health services (45). They remain, therefore, to blend into health policies, with financial and logistical support to the municipalities, associated with the attraction of health professionals as they present greater efficiency and effectiveness in the collective health outcomes (52).

Our results agree with the studies reviewed, related to the importance of retaining human resources and establishing the best organizational models in PHC in order to gain equity (in access and quality) and efficiency. The models that take into account the geographical distances and local environmental characteristics proved to be more efficient. The farther the locality, the more mobile health teams and facilities appear to be the best alternative.

We have reported several limitations in our study along the way, either methodological (aggregation of data) or in interpretations (symbolic aspects of place), always seeking to overcome these difficulties with the lessons learned from our experience. We still lack more specific variables that may describe intersectional aspects more clearly, as well as customized measures for remote rural localities that may surpass the limits of the health production model (53). Our main strength is to study efficiency simultaneously with effectiveness through an equity lens that broadens our view and reduces distortions, filling an important gap in the literature.

The main limitation of our study regards the use of secondary/aggregated data created for different purposes, coming from different databases and timeframes. We have treated the data accordingly, with control of time invariant characteristics, with the fixed effects models.

Future research should delve with a mixed methods frame, with more contextual information and capacity to build more robust indicators that capture the local reality, with primary data, allowing measuring more directly the efficiency/effectiveness of local health systems.

5 Conclusion

Brazilian states with RRL show superior resource and health efficiency through utilization of services according to health needs. These models showed strong determination and the final models included intersectional equity. The main message is that reducing intersectional inequities in income and education by ethnicity could greatly increase the efficient attainment of health levels in society. The remote rural typology demonstrated greater efficiency and effectiveness in health than the other typologies in the states containing RRL. The organizational models with the FHS teams and the CHW visits played a key role, together with *per capita* health expenditures and intergovernmental transfers. Among the remote rural municipalities, the Amazon region clusters (North Roads/Waters) stand out, with models presenting variables relevant to local reality, such as the coverage of fluvial and riverine teams, and especially intersectional gender and ethnicity equity in education.

Hopefully, our findings shall contribute to gains in efficiency and effectiveness in these spaces, through health policies prioritizing financial resources (local and from other levels of government) and the settlement of health professionals, family health teams configured for the necessary displacements, the availability of local diagnosis and treatment, and closer attention to basic sanitation and intersectional inequities in education. Finally, we must aim for equity of gender and ethnicity in income and education, and above all of place, perceived in its entirety.

Data availability statement

Datasheets are also available as Supplementary materials. The original contributions presented in the study are included in the article/Supplementary material, further inquiries can be directed to the corresponding author. The consulted data sources are open to public access and can be reached at the following websites: IBGE—Brazilian Institute of Geography and Statistics (demographic and health surveys), https://www.ibge.gov.br/estatisticas/sociais/populacao/9662-censo-demografico-2010.html?=&t=o-que-e, https://www.ibge.gov.br/estatisticas/sociais/saude/9160-pesquisa-nacional-de-saude.html; DATASUS—SUS Information Technology Department (CIH, CNES, SIA, SIM, SINAN, SINASC; SIAB; SISMAMA; SISCOLO), https://datasus.saude.gov.br/informacoes-de-saude-tabnet/; e-gestor AB—e-manager-PHC, https://acesso-egestoraps.saude.gov.br/login; MDS—Social Development Ministry, https://legado.dados.gov.br/dataset?_license_id_limit=0&organization=ministerio-do-desenvolvimento-social-mds&res_format=JSON&license_id=cc-by&tags=mi+social&_res_format_limit=0; MPF—Federal Prosecution Ministry, http://combateacorrupcao.mpf.mp.br/ranking; SIOPS—Public Health Budget

Information System, http://siops-asp.datasus.gov.br/cgi/siops/serhist/MUNICIPIO/indicadores.HTM; STN/MF—National Treasure Department/Ministry of Finance, https://www.gov.br/tesouronacional/pt-br; UNDP—United Nations Development Program, https://www.undp.org/pt/brazil/idhm-munic%C3%ADpios-2010.

Ethics statement

Obtaining ethic approval is not applicable for this postdoctoral project, as publicly available data was used in this study. The main research, which this project relates to, was approved by the Ethics Committee of the Faculty of Public Health of the University of São Paulo under CAAE number 37672620.8.0000.5421, statement 4.285.824, as of September, 18th, 2020.

Author contributions

SS: Conceptualization, Data curation, Formal analysis, Funding acquisition, Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. AB: Conceptualization, Funding acquisition, 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.2024.1401193/full#supplementary-material

SUPPLEMENT 1
Statistical techniques in more detail (DEA and FE).

SUPPLEMENT 2
Rural-urban classifications and typologies (IBGE).

SUPPLEMENT 3
Typologies of the Brazilian remote rural localities.

SUPPLEMENT 4
Bivariate efficiency analysis by dimension-all Brazilian states analysis.

SUPPLEMENT 5
Graphs by macro-regions and Federative Units-all Brazilian states efficiency analysis and more details about the Brazilian states.

SUPPLEMENT 6
Bivariate analysis by dimension - Brazilian states with RRL only.

SUPPLEMENT 7
Graphs by rural-urban typologies for Brazilian states with RRL only. Ranking of the Brazilian states, according to rural-urban typology.

SUPPLEMENT 8
Summary chart of variables in the final models for the three analysis and metadata.

SUPPLEMENT 9-12
Datasheets with information on the three analyses performed.

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EDITED BY
María Del Carmen Valls Martínez,
University of Almeria, Spain

REVIEWED BY
Henrik Lerner,
Marie Cederschiöld University, Sweden
Ming Shao,
Beijing Forestry University, China

*CORRESPONDENCE
Alexander Welker Biondo
✉ abiondo@ufpr.br
Alessandra Cristiane Sibim
✉ alessandra.sibim@ufpr.br
Wagner Antonio Chiba de Castro
✉ wagner.chiba@gmail.com

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One Health Index applied to countries in South America

Alessandra Cristiane Sibim[1,2]*,
Wagner Antonio Chiba de Castro[3]*, Louise Bach Kmetiuk[4]
and Alexander Welker Biondo[2]*

[1]Latin-American Institute of Technology, Infrastructure and Territory, Federal University for Latin American Integration (UNILA), Foz do Iguaçu, Brazil, [2]Graduate College of Veterinary Sciences, Department of Veterinary Medicine, Federal University of Paraná State, Curitiba, Brazil, [3]Latin-American Institute of Life and Nature Sciences, Federal University for Latin American Integration (UNILA), Foz do Iguaçu, Brazil, [4]Zoonoses Surveillance Unit, Municipal Secretary of Health, Curitiba, Brazil

Introduction: The One Health concept has proposed an integrated and unified approach aiming for health balance and enhancement by recognizing the interdependence of human, animal, and environmental health. The COVID-19 pandemic has pushed global One Health initiatives and policy improvement toward preventive measures for future pandemics, particularly of zoonotic origin. Such a scenario may be particularly relevant for South America, which is considered highly vulnerable due to its natural biodiversity superposed to socioeconomic and environmental issues, demanding effective methods and indicators for proper One Health strategies and goals that are aligned with macroregional contexts.

Methods: Accordingly, the present study aimed to assess the One Health Index (OHI) in South American countries, along with potential interactions with socioeconomic indicators. The results obtained using clustering analysis and permutational multivariate analysis of variance (PERMANOVA) have revealed a positive association between the OHI and the Human Development Index (HDI) but not with gross domestic product (GDP).

Results: Although South American countries with political stability, robust investment in health, and progressive policies have shown a higher OHI, better environmental health is not associated with better human and animal health. In addition, although the Amazon biome— spanning 9 of the 12 South American countries—has positively impacted environmental health, this benefit contrasts with the rudimentary local human health systems, highlighting the complexity of One Health within the South American context. The lack of stronger indicators for animal health was also considered an important weak point for a true OHI assessment. Nonetheless, countries with more developed livestock have presented better animal health, which may not reflect an overall animal health indicator, as companion and wildlife animal health indicators were not available.

Discussion: Although lower (within-country) scale analysis such as states and metropolitan areas may better shape internal differences, the study herein has clearly shown One Health inequalities and challenges among South American countries. Equally important, forests and other natural areas in developing countries, particularly the Amazon, should receive incentives to promote sustainable economic growth. This approach would help prevent sacrificing environmental health for the benefit of human and livestock animal health.

KEYWORDS

health indicators, socioeconomical factors, ecosystemic services, livestock, political stability

1 Introduction

One Health has been defined as an integrated and unified approach aiming for a sustainable assessment of human, animal, and environmental health, with a holistic strategy existing long before the term was coined (1). A recent One Health consensus report has considered humans, domestic and wildlife animals, plants, and their ecosystemic environment as intimately connected and interdependent (2), with multidisciplinary and professional integration aiming to better recognize zoonotic emergences from a One Health perspective (3).

The recent COVID-19 pandemic has highlighted the need for a quadripartite agreement to support such initiatives (4). This agreement emphasizes preventing future epidemics through a global understanding of new disease emergence (5) and has been officially approved by the Pan-American Health Organization (PAHO) as a tool for dealing with health threats at the human–animal interface (6). The first action point of this policy resolution aimed to perform an analysis and map the complex interactions among actors and processes in the fields of human, animal, plant, and environmental health in such specific national contexts.

South America has been considered the Americas meridional portion, including 12 sovereign states and two dependent territories (7). Although comprising some of the most important natural reserves worldwide, such as the Amazon Rainforest and Andes, several cultural heritage sites, and large cultivable areas, the South American region remains mostly underdeveloped and highly vulnerable to deforestation, poaching, and zoonotic diseases (8). Although nine South American countries share the Amazon forest, the world's largest tropical rainforest (9), which provides a significant carbon sink service and helps regulate global climate (10), several socioeconomic and environmental threats (11, 12) have demanded a trans-and multidisciplinary One Health approach to fight such complex challenges (13).

Although widely recognized, the One Health approach has required better and adapted strategies for specific socio-ecological contexts (14, 15). This includes the integration of information technology and statistical analysis to assess environmental and sustainable effectiveness (16), improved climate and environmental comprehension (17), evaluation of risk from current and future natural disasters (18), identification of vulnerable populations and areas (19), and the practical use of the One Health Index itself (20). Accordingly, the present study aimed to assess available indicators of human, animal, and environmental health from a statistical and comparative One Health Index (OHI) perspective of all 12 South American countries.

2 Materials and methods

2.1 Countries and socioeconomic factors

The present study included all 12 South American countries, namely Argentina, Bolivia, Brazil, Chile, Colombia, Ecuador, Guiana, Paraguay, Peru, Suriname, Uruguay, and Venezuela. The socioeconomic factors of these countries were obtained from official reports and sites and included the Human Development Index (HDI) (21) and the gross domestic product (GDP) *per capita*, based on the purchasing power parity (PPP) (22). The Human Development Index (HDI) has been described as a measure of a country's well-being and development, combining life expectancy, education, and standard of living, as the geometric mean of normalized indexes for each of these three dimensions (23). The gross domestic product (GDP) *per capita*, based on purchasing power parity (PPP), represents the average value of all goods and services produced by a country in a given year, adjusted for price differences, and expressed in international dollars, providing a more accurate measure of a country's comparative living standard (24).

2.2 Human, animal, and environmental health indicators

Updated indicators for human, animal, and environmental health of all 12 South American countries were selected from the available literature and official reports and sites to construct the One Health Index (Table 1; Supplementary Tables 1–4), based on the One Health Index previously established by our research group (20).

2.3 Data analysis and One Health Index construction

The assessment of each country was based on nine indicators, which were equally distributed into three indicators per health category (Table 1). A performance (ranking) score was attributed to each country for each indicator, with the lowest graded as 1, the highest graded as 12, and the remaining ones graded accordingly (Table 2). The final grade for each country in each health category

TABLE 1 Indicators for human, animal, and environmental health to construct the One Health Index of all 12 South American countries.

Health category	Performance indicators (PI)	Source	Reference
Human	1. GHS Index	Global Health Security Index	(44)
	2. Social Vulnerability	Multidimensional Vulnerability Index	(29)
	3. Vulnerability	World Risk Report	(45)
Animal	4. Zoonoses	Global Health Security Index	(44)
	5. Pesticides	Food and Agriculture Statistics	(46)
	6. WAHIS*	World Organisation for Animal Health	(40)
Environmental	7. Environmental vulnerability	Multidimensional Vulnerability Index	(29)
	8. Vulnerability to climate changes	Universal Vulnerability Index	(47)
	9. Environmental performance	Environmental Performance Index	(16)

*Qualitative information parameters (absence and presence) of each country were divided into: 1. Disease; 2. serotype, subtype, and genotype; 3. animal category; 4. outbreak identifier; and 5. vaccinated.

was the average of the three indicators. Thus, each sampling unit was the composite result of indicators, representing the three weighted grades, corresponding to each of the three health categories, as established and adapted (20). The One Health Index (OHI) of each country was calculated as the average of grades from the three correspondent health categories. It is important to mention that such applied methodology has resulted in composite indexes reflecting a relative panorama of One Health among South American countries, with a comparative rather than absolute OHI.

2.4 Ranking (score) according to socioeconomic factors

Countries were ordered according to their grades of each health category and analyzed using principal component analysis (PCA), which classified countries using cluster analysis, exploring 1. One Health Index, 2. Human Development Index (HDI), and 3 gross domestic product (GDP) *per capita*, based on purchasing power parity. Statistical significance for country clusters in each factor was assessed using clustering analysis and permutational multivariate analysis of variance (PERMANOVA) (25), based on the three first coefficients of principal components, obtained from each PCA. A *p*-value less than 0.05 was considered significant. All statistical analyses were performed in the statistical environment R (26).

3 Results

The results of One Health for each South American country were obtained, gathered, and presented (Figure 1; Supplementary Table 5). Overall, Uruguay (8.0), Chile (7.9), and Argentina (7.6) presented the highest grade of One Health. Guiana (3.7), Bolivia (5.2), and Venezuela (5.4) presented the lowest grade of One Health. The principal component analysis, presented in Figure 2, illustrates the significant differences among South American countries based on human, animal, and environmental health categories, as determined by the One Health grading (PERMANOVA; $F = 3.6305$; $p = 0.009$). Countries with higher One Health grades (Uruguay, Chile, and Argentina) were grouped to the left of the graphic.

The same pattern can be observed in significant differences among countries on health categories compared to the Human Development Index (HDI) (Figure 3). Chile (0.855), Argentina (0.842), and Uruguay (0.809) (countries grouped to the left of the graphic) also presented higher HDI (PERMANOVA; $F = 4.9113$; $p = 0.001$). However, no significant difference was observed for South American countries and gross domestic product (GDP) *per capita* based on purchasing power parity (PPP) (PERMANOVA; $F = 2.0781$; $p = 0.10$) (Figure 4). Guyana presented, at the same time, the highest GDP-PPP (U$ 60,650) and the lowest OHI (3.7).

4 Discussion

The results indicated that South American countries with higher Human Development Indexes (HDIs) also had higher One Health Indexes (OHIs), which are composed of various education and health indicators. However, the results also showed no significant association between the One Health Index and gross domestic product (GDP) *per capita*. This suggests that the holistic nature of One Health is better explained by the HDI's social approach rather than by an economic index such as GDP, even when adjusted for purchasing power. Thus, despite the importance of explaining health in several contexts (27, 28), the economic factor was not the sole determinant of One Health.

The study herein showed that South American countries with higher environmental health also presented lower human and animal health. While Ecuador (1st), Colombia (2nd), Suriname (3rd), and Venezuela (4th) presented the highest scores in environmental health, their performance in animal health (Ecuador 9th and Suriname 11th) and human health (Colombia 10th, Ecuador 11th, and Venezuela 12th) was relatively lower. The environmental health indicators explored aspects such as climate change mitigation, air quality, biodiversity and ecosystemic services,

TABLE 2 Performance (ranking) grades attributed to each South American country, based on the nine performance indicators (PI), which comprise the environmental, animal, and human health categories, along with the grades for each category, expressed as a total.

Countries	Environmental health				Animal health				Human health			
	PI.1	PI.2	PI.3	Total	PI.4	PI.5	PI.6	Total	PI.7	PI.8	PI.9	Total
Argentina	3	1	6	3.3	9	10	8	9.0	10	11	10	10.3
Bolivia	4	4	4	4.0	5	4	10	6.3	2	7	7	5.3
Brazil	6	8	8	7.3	11	12	4	9.0	8	5	4	5.7
Chile	2	2	12	5.3	4	9	7	6.7	12	12	11	11.7
Colombia	11	10	7	9.3	10	11	1	7.3	9	4	1	4.7
Ecuador	12	11	11	11.3	8	8	2	6.0	7	1	3	3.7
Guiana	1	7	2	3.3	1	2	3	2.0	3	6	8	5.7
Paraguay	5	5	5	5.0	6	6	9	7.0	5	2	9	5.3
Peru	9	3	3	5.0	7	7	11	8.3	11	9	2	7.3
Suriname	10	9	9	9.3	3	1	5	3.0	4	8	6	6.0
Uruguay	8	12	1	7.0	12	5	6	7.7	6	10	12	9.3
Venezuela	7	6	10	7.7	2	3	12	5.7	1	3	5	3.0

fishing, and water resources (16), as well as vulnerability to natural disasters (29) and risk areas (30). These South American countries have their territory overlapped by the Amazon forest (42% of Colombia, 48% of Ecuador, and 94% of Suriname), with most populations living in close contact with natural areas (31). Thus, the ecosystemic services provided by the rainforest may have a favorable impact on applied performance indicators of environmental health. The Amazon forest has been threatened by illegal human activities such as logging, mining, and fires. Notably, the southern and southeastern Brazilian regions of the Amazon have experienced increasing soil erosion and a 7% deforestation rate (411,857 km^2) between 1960 and 2019, due to expanding agriculture and livestock activities (32). The annual Brazilian Amazon deforestation has surpassed 13,000 km^2 from 2019 to 2021, which represents an increase of 56.6% when compared to 2016–2018 (33). Such an increase was reportedly associated with a government attempt to promote environmental sustainability through the agribusiness-based economy in the southern and southeastern Brazilian Amazon, which culminated in land grabbing, conflicts, and deforestation (33). In addition, modeling studies have indicated an increase of 4°C in temperature or deforestation exceeding 40% as two "tipping points" of irreversible changes for biodiversity and ecosystems of the Amazon forest (34). In such a scenario, recent studies have advocated for sustainable development in the Amazon, based on the non-use of natural resources, accompanied by an effort to improve ecosystem resilience (34, 35). Thus, considering the environmental health role for a better One Health, forests and other natural areas of developing countries (particularly the Amazon forest) should receive incentives for sustainable economic growth, preventing the sacrifice of environmental health for the benefit of human and livestock animal health. Although the data presented reflect the most recently available information, providing only a current temporal snapshot, the historical overall development situation in South America and its negative impact on the Amazon Rainforest as a side effect consequence over time should be considered a warning for a truly sustainable and healthy development of the region.

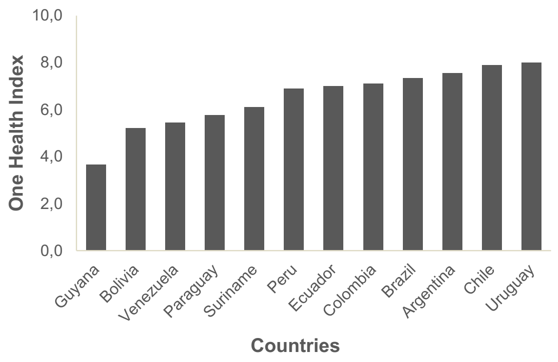

FIGURE 1
Graphic of One Health Index grading of South American countries.

Despite having large natural areas, South American countries have deep health system limitations, with recent trajectories of health

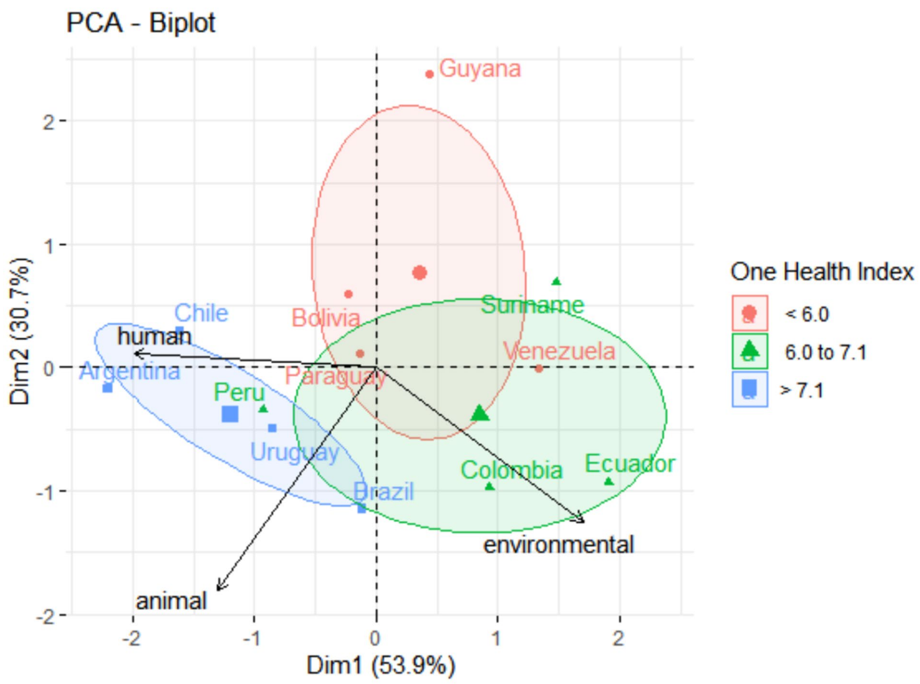

FIGURE 2
Graphic of principal components analysis (PCA) showing the influence of (human, animal, and environmental) health variables on all the South American countries. Colors and ellipses circling the country groups represent the confidence ellipsis, which delimited country clusters according to grading intervals of One Health.

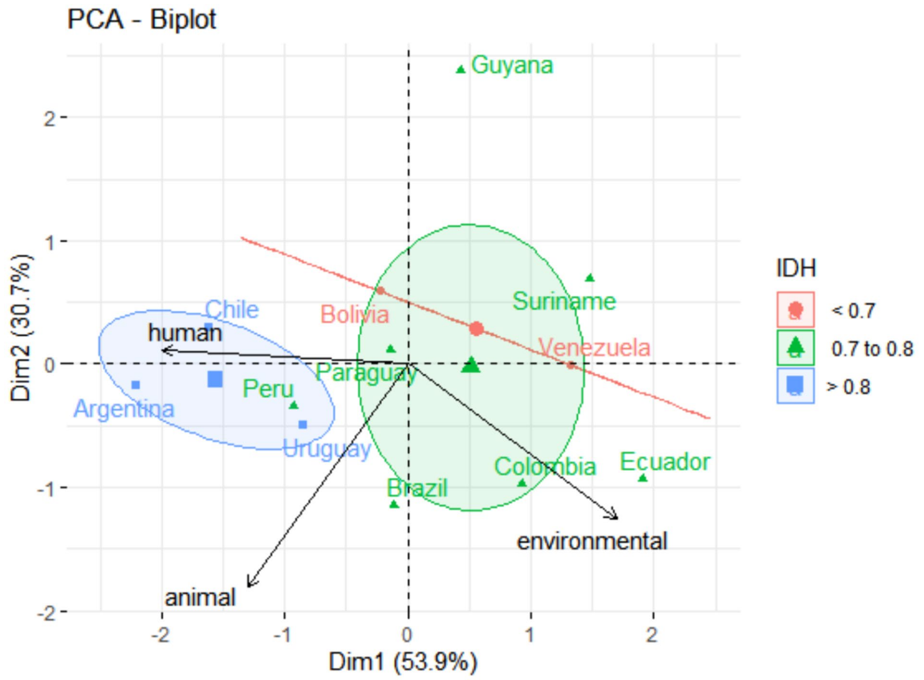

FIGURE 3
Graphic of principal component analysis (PCA) showing the influence of (human, animal, and environmental) health variables on all the South American countries. Colors and ellipses circling the country groups represent the confidence ellipsis, which delimited country clusters according to the higher Human Development Indexes (HDIs).

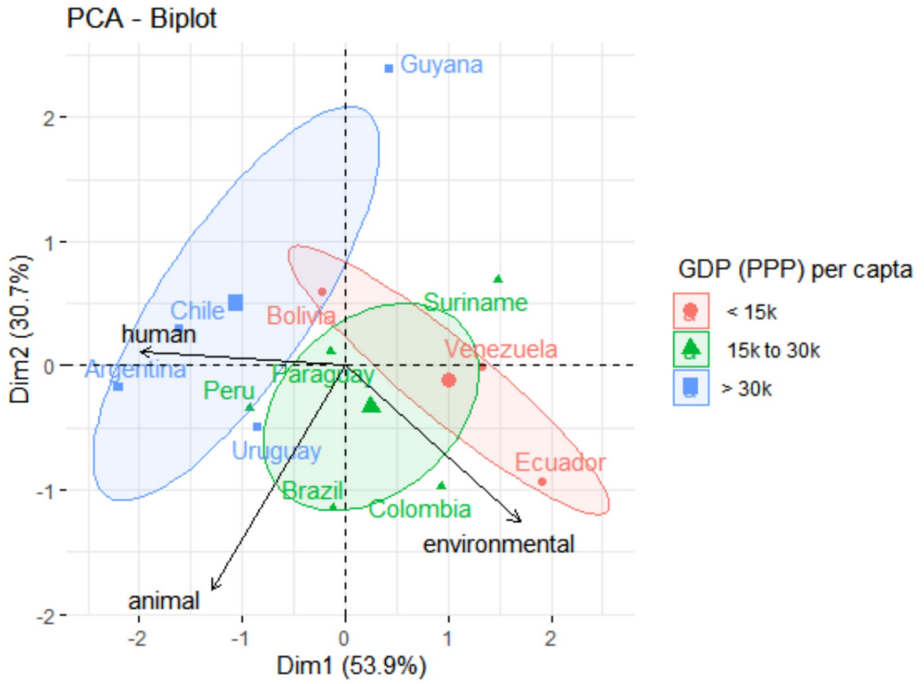

FIGURE 4
Graphic of principal component analysis (PCA) showing the influence of (human, animal, and environmental) health variables on the South American countries. Colors and ellipses circling the country groups represent the confidence ellipses, which delimited country clusters according to the gross domestic product (GDP) *per capita*, based on purchasing power parity (in U$).

privatization and increased access inequalities to health services (36), political crises affecting the quality of services provided (37), and shortage of human resources (38). Thus, South American countries with political stability, higher investment in human health, and progressive political characteristics have been placed at a higher level of the One Health Index (OHI). The four countries with the highest scores in the OHI (Uruguay, Chile, Argentina, and Brazil) were among the five countries with the highest investments in human health *per capita* (39), presenting solid democratic political regimes throughout the last decades.

A limitation of this study is the difficulty in accessing animal health indicators, which were used along with several standard composite indexes of human and environmental health. Only one integrated animal health index was found at the global level, the Animal Protection Index (API), recently provided by the World Animal Protection (WAP), a non-profit organization. However, such an index was not used herein, as only 7 of 12 (58.3%) South American countries presented available API grades. Thus, only indirect performance indicators of animal health were explored, such as zoonoses, pesticides (harmful to natural biota), and a selection of livestock indicators obtained from the World Animal Health Information Systems (WAHIS), a database maintained by the World Organisation of Animal Health (40). Thus, the animal health approach was based exclusively on livestock health (and not welfare), excluding analysis of both companion and wildlife animal health.

A previous One Health Index (OHI) study conducted at the city level Curitiba, the eighth biggest metropolitan area of Brazil, has also shown difficulties in obtaining animal health indicators (20, 41). In this study, qualitative (yes or no) indicators were used, assessing only companion animal health, including education and neutering/spaying programs, animal hoarder monitoring, enforcement against animal cruelty, microchipping, and adoption of abandoned pets. Such a lack of comprehensive and reliable data in the present study may have biased animal health as livestock health only. Thus, indicators may have rewarded agricultural performance, such as control of animal diseases and conscient use of pesticides. In such a scenario, South American countries with advanced livestock production, such as Brazil, Argentina, and Uruguay, were among the highest scorers for animal health in South America. Thus, further efforts and studies should focus on providing reliable animal health indexes for livestock, companion animals, and wildlife, which could then be used for comparisons at city, state, country, and continental levels.

As another limitation, the present study has assessed information at international official organizations, at country level, available in official languages of South American countries including English (Guiana), French (French Guiana), Portuguese (Brazil), and Spanish (all others), with exception of Dutch in Suriname and official native indigenous languages such as Guarani in Bolivia and Paraguay, Aymara in Bolivia and Peru, and Quechua in Bolivia, Ecuador and Peru. In addition, information obtained, particularly in large territorial countries, may not represent the within-country inequalities among states and provinces, or even among cities within the same state or province. In addition, as well-known, the largest South American metropolitan areas have been characterized by deep within-city inequalities, such as São Paulo, Buenos Aires, and Santiago cities (42). As the One Health Index applied at the country level may be impaired by inequalities and disparities at the state and city levels, further studies should compare and contrast One Health Index patterns across local, metropolitan, and regional regions within countries.

Despite the authors' recognition of the importance of temporal and spatial analyses, the data herein did not support a temporal analysis because the surveys only included the most recent available data, providing a current temporal snapshot. Although the data herein did not support spatial analysis, the discussion was focused on countries overlying the Amazon rainforest biome, which is important for current analysis and further research. The authors also acknowledge that the data herein may be insufficient to explore smaller spatial scales (such as large metropolitan areas), which would enhance the understanding of the One Health landscape in South America.

Finally, although in the highest available resolution and with bigger letters and captions, the figures presented in the present study have a standard outcome layout provided by the Tidyverse, Stats, and Factoextra statistical packages, which were used to build them.

Other One Health assessments have been recently reported, focusing on interdisciplinary setting effectiveness, with assessment tools of a calculated hexagon presented as OH-index (OHI) and OH-ratio (OHR) in spider diagrams, along with Theory of Change (TOC) as indicator for measurement of expected results, comparing One Health with conventional health initiatives (43). In addition, a Global One Health Index (GOHI) based on a three-layer framework has been proposed for the evaluation of One Health structure, process, and outcome (51). Although presenting important contributions to One Health assessment, both studies may lack the practical approach presented by the One Health Index (OHI) applied herein (20), which has combined several health indexes within human, animal, and environmental components, providing holistic and comparative strengths and weaknesses among municipalities.

The holistic One Health Index (OHI) approach herein has provided a better understanding of health as a whole in South American countries, contextualized by the Human Development Index (HDI) and contrasted by the gross domestic product (GDP) *per capita*. The higher grades of environmental health in some South American countries have not necessarily indicated better human and animal health. Progressive policies, consistent investments in human health, and political stability were important factors associated with higher One Health grades. The limitations and lack of reliable indicators, particularly for environmental and animal health, have highlighted the need for better indexes worldwide. Although within-country inequalities may have influenced the results of the present study, this is the first attempt to compare One Health in such a practical manner, and further studies should address local, metropolitan, and regional regions within countries in South America and other continents.

Data availability statement

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

Author contributions

AS: Conceptualization, Formal analysis, Investigation, Methodology, Software, Validation, Visualization, Writing – original draft, Writing – review & editing. WC: Conceptualization, Formal analysis, Investigation, Methodology, Software, Validation, Visualization, Writing – original draft, Writing – review & editing. LK: Investigation, Methodology, Writing – review & editing. AB: Conceptualization, Methodology, Visualization, Writing – review & editing.

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Conflict of interest

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

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1394118/full#supplementary-material.

SUPPLYMENTARY TABLE 1
Socioeconomic data of the countries, represented by the Human Development Index (HDI) (23) and the Gross Domestic Product per capita adjusted by purchasing power parity (in dollars) (48) of South American countries, as well as the considered classes.

SUPPLYMENTARY TABLE 2
Raw values of the environmental health performance parameters of South American countries. EV_MVI = Environmental Vulnerability, Multidimensional Vulnerability Index (UN, 2023); PVCCI_UVI = Physical Vulnerability to Climate Change Index, Universal Vulnerability Index (49); EPI = Environmental Performance Index (50).

SUPPLYMENTARY TABLE 3
Raw values of the animal health performance parameters of South American countries. ZD_GHS = Zoonosis, Global Health Security Index (Bell and Nuzzo, 2021); PEST_FAO = Pesticides (kg*ha-1), Food and Agriculture Statistics; WAHIS = Parameters, World Organisation for Animal Health.

SUPPLYMENTARY TABLE 4
Raw values of the human health performance parameters of South American countries. SV_MVI = Social Vulnerability, Multidimensional Vulnerability Index; VUL_WRR = Vulnerability, World Risk Report (51); GHS = Global Health Security Index.

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EDITED BY
Olatunde Aremu,
Birmingham City University, United Kingdom

REVIEWED BY
Jiang Chen,
Zhejiang University, China
Zhengbin Zhang,
Wuhan Pulmonary Hospital, China

*CORRESPONDENCE
Zhaoyan Chen
✉ chenzhaoyan1993@163.com

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Modification of gemcitabine with oxaliplatin in China for unresectable gallbladder cancer: a cost-effectiveness analysis

Zhaoyan Chen* and Fangyuan Tian

Department of Pharmacy, National Clinical Research Center for Geriatrics, West China Hospital, Sichuan University, Chengdu, Sichuan, China

Background: The incidence of gall bladder cancer (GBC), one of the most prevalent bile duct malignancies, differs with ethnicity and geographic location. To treat unresected GBC in the Chinese setting, this study aimed to assess the financial effectiveness of a combination of modified gemcitabine and oxaliplatin.

Methods: Data from a randomized controlled study in which individuals with metastatic GBC were treated with oxaliplatin and gemcitabine demonstrated improved survival. A Markov model is built to calculate the incremental cost–benefit ratio (ICER) from the viewpoint of Chinese society on the basis of clinical symptoms and disease development. One-way certainty and probability sensitivity analyses are used to describe the uncertainty in the model.

Results: Compared with those of fluorouracil (FU) and folinic acid, the utility value of modified oxaliplatin combined with gemcitabine increased by 0.22QALY throughout the course of the 10-year simulation (FA). In a Chinese healthcare setting, the cost-effectiveness ratio (ICER) is $52765.59/QALY, with a 0% chance of cost–benefit at the WTP (willing-to-pay) level of $37697.00/QALY. The ICERs predicted by sensitivity analysis were not significantly affected by cost variations related to the management of Grade 3–4 AEs, the diagnostics used, or hospitalization expenditures.

Conclusion: In a Chinese healthcare context, modified gemcitabine coupled with oxaliplatin (mGEMOX) is not a cost-effective treatment option for unresectable GBC.

KEYWORDS

gallbladder cancer (GBC), gemcitabine, oxaliplatin, fluorouracil, cost effectiveness

Introduction

Fewer than 5,000 new instances of gallbladder cancer (GBC) are detected in the United States each year, and rates vary by geographic region and race (1). For GBC, surgery is the sole treatment option. However, only a small number of patients are suitable for curative surgery, with the remainder receiving palliative care, due to the extent of the lesion (including locally advanced unresectable lesions due to local invasion of critical structures or lesions that metastasized beyond local regional boundaries) (2). Currently, there is no standard chemotherapy for treating GBC (3, 4). Gemcitabine and oxaliplatin have demonstrated effects on the biliary tract in patients with pancreatitis and GBC either alone or in combination with other treatments (5–7). The third-generation platinum drug oxaliplatin is substantially less nephrotoxic and emetic than large doses of cisplatin. A good substitute for gemcitabine and cisplatin may be modified gemcitabine coupled with oxaliplatin.

According to recent findings from a phase III single-center trial conducted in India, the combination of oxaliplatin and gemcitabine significantly prolonged median progression-free survival (PFS) ($p < 0.001$) and median overall survival (in months) ($p = 0.039$) in patients with unresectable GBC (8). Data on the top-selling pharmaceuticals worldwide indicate that oxaliplatin generated nearly $20 billion in sales over the 20-year period from 1999--2019. The cost of oxaliplatin has decreased somewhat in China due to escalating competition from generic medications. In Sichuan Province, a tube of oxaliplatin (50 mg) costs US$326.59. However, the cost-effectiveness of a pharmacological treatment plan is one of the factors that influences the ultimate selection in a nation such as China, which has inadequate medical resources. The potential cost advantages of the mGEMOX regimen for the treatment of patients with unresectable GBC were assessed in this study via a Markov model.

Materials and methods

Target population

Patients who were 18 years of age or older with unresectable GBC verified by biopsy or fine needle aspiration cytology met the inclusion criteria. If a patient has previously received adjuvant chemotherapy and/or radiotherapy, it should be completed at least 6 months before recruitment into this study. Everyone in the PFS health status group first received one of the two treatments. For a maximum of 6 cycles or until intolerable toxicity, whichever came first, the patients in the intervention group received 900 mg/m^2 gemcitabine and 80 mg/m2 IV infusion (mGEMOX) oxaliplatin on days 1 and 8 of every 3 weeks. Patients in the comparison group received an intravenous bolus of FA 20 mg/m^2 and FU 425 mg/m^2 once a week for 30 weeks (FUFA).

Model structure

The cost-effectiveness of the two treatment modalities was compared via the Markov decision tree model. Progression-free survival (PFS), progressive disease (PD), and death are the three mutually exclusive states included in the model. All patients with metastatic or unresectable GBC begin therapy for PFS; however, they may transition over time to other health states (Figure 1). Patients in the PFS state may enter PD or death states after a Markov cycle, or they may remain in the PFS state. Patients in the PD state, however, are unable to return to the PFS condition, and any patient may enter the death state. An absorbed state is the death state. Toxicology tests were performed on all patients. Toxicology was determined via the National Cancer Institute Common Toxicity Criteria (version 3.0). The model timeframe is set to 1 month on the basis of clinical symptoms and the rate of disease progression. The following formula was used to convert monthly transition probabilities from median survival estimates (Table 1): the formula $p = 1-e-R$, where $R = -\ln[0.5]/$(time to incident/number of treatment cycles), was used to obtain P (1 month) = 1-(0.5, 1/median time to event), which was then used to calculate P (1 month) (9, 10). For this model, a 10-year time horizon was selected.

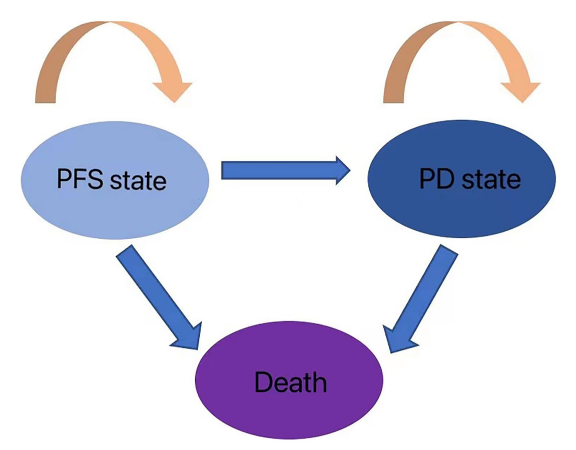

FIGURE 1
Markov model health states and transition rules.

TABLE 1 Transition probabilities between unresectable GBC states.

Transition probabilities	Baseline value	Lower limit	Upper limit
mGEMOX			
$P_{pfs\text{-}pfs\text{-}1}$	0.85	0.68	1.00
$P_{pfs\text{-}pd\text{-}1}$	0.08	0.06	0.09
$P_{pfs\text{-}death\text{-}1}$	0.07	0.06	0.08
$P_{pd\text{-}pd\text{-}1}$	0.50	0.40	0.60
$P_{pd\text{-}death\text{-}1}$	0.50	0.40	0.60
FUFA			
$P_{pfs\text{-}pfs\text{-}2}$	0.68	0.54	0.82
$P_{pfs\text{-}pd\text{-}2}$	0.18	0.14	0.22
$P_{pfs\text{-}death\text{-}2}$	0.14	0.11	0.17
$P_{pd\text{-}pd\text{-}2}$	0.53	0.43	0.64
$P_{pd\text{-}death\text{-}2}$	0.47	0.37	0.56

FUFA, fluorouracil and folinic acid; mGEMOX, modified gemcitabine and oxaliplatin; PFS, progression-free survival; PD, progressive disease.

Model parameters

Expenses were calculated with patient payments in mind (Table 2). Anticancer medications, diagnostics (total abdominal enhanced CT, biochemical examination), management of grade 3–4 adverse events (AEs), and hospitalization expenditures were taken into account during the analysis. Individual differences resulted in implicit costs being disregarded. We hypothesized that the average patient would weigh 65 kg, stand 1.64 m tall, and have a BSA of 1.72 m^2 (11). The 2023 charge standards of West China Hospital, Sichuan University, were consulted to determine the unit price for each medication and test. We assessed the cost of second-line treatment in the two groups on the basis of the data from the trial by Sharma et al. (8) and the progression of the disease. The conversion

TABLE 2 Model parameters related to cost and effectiveness in patients with GBC.

Parameters	mGEMOX	FUFA
Clinical efficacy, months		
Median PFS, months	8.5	3.5
Median OS, months	9.5	4.6
Probability of grades 3–4 adverse events, %		
Vomiting	7.69	7.14
Myelosuppression	38.46	7.14
Neurotoxicity	11.54	0
Transaminitis	15.38	0
Neutropenic fever	7.69	0
Unit costs, $/months		
Cost of tests	90.03	164.37
Hospitalization	140.76	164.22
Cost for adverse events	1.26	/
Cost for the progressive disease state	2017.94	1834.49
Annual discount rate, %	5	5

FUFA, fluorouracil and folinic acid; mGEMOX, modified gemcitabine and oxaliplatin; PFS, progression-free survival; PD, progressive disease.

rate used to convert all costs to US dollars was $1 = 6.82 (average exchange rate for January 2023) (12). This model uses health outcome data from single-center, randomized, controlled, open-label research. Years of quality-adjusted life are used to express the health utility value (QALY). Given that fundamental utility information is absent from the original literature, the health utility value is derived from published literature (13). The utility values for death status, progressive disease, and progression-free disease are 0.77, 0.64, and 0.00, respectively. Table 2 displays the model parameters for cost and effectiveness. The cost and utility value are both reduced at a 5% annual rate in accordance with the "China guidelines for pharmacoeconomic evaluations."

Sensitivity analyses

The findings of the deterministic one-way sensitivity analysis revealed variation in all the parameters utilized in the evaluation (with the exception of the discount rate; range = 20%). The discount percentage varied from 0 to 8%. A second-order Monte Carlo simulation was used to perform a probabilistic sensitivity analysis over the course of 1,000 iterations. To assess the most successful techniques at different willingness-to-pay (WTP) thresholds, cost-effectiveness acceptability analysis was performed. The WTP benchmark was set at three times China's $37697.00/QALY GDP *per capita* in 2022.

Results

Base-case analysis

The findings of the 10-year simulation demonstrate that the utility value of modified oxaliplatin in combination with gemcitabine is increased by 0.22QALY (0.45QALY vs. 0.23QALY) compared with that of FU and FA (Table 3). Similar to the cost increase, the cost-effectiveness ratio (ICER) shows a 0% chance of being cost-effective at the WTP level of $37697.00/QALY, and the cost increase is 11608.43 US dollars per person (Figure 2). Given its overall cost implications, this combination is not a cost-effective treatment option for unresectable gallbladder cancer. For patients in the PFS illness state, the cost of the mGEMOX regimen is more than 10 times greater than the cost of the FUFA regimen ($12790.39 vs. $1121.48).

TABLE 3 The results of the cost-effectiveness analysis.

Parameters	mGEMOX	FUFA
Costs for the PFS state, $	12790.39	1121.48
Costs for the PD state, $	2111.92	2172.40
Total costs, $	14902.31	3293.88
Incremental costs, $	11608.43	/
Effectiveness for the PFS state, QALYs	0.39	0.17
Effectiveness for the PD state, QALYs	0.06	0.06
Total effectiveness, QALYs	0.45	0.23
Incremental effectiveness, QALYs	0.22	/
Total C/E, $/QALY	33116.24	14321.22
ICER, $/QALY	52765.59	/

Sensitivity analysis

To evaluate the effects of specific Markov model parameters, a one-way sensitivity analysis was performed. An illustration of the outcomes is shown in a tornado diagram (Figure 3). The most important model parameters were the cost of oxaliplatin for the mGEMOX group, the utility of the PFS state, and the cost of the PFS state, all of which exhibited a variance of approximately 20%. The ICER increased from $42068.81/QALY to $65850.30/QALY as the PFS state cost changed from $1684.68/month to $2527.02/month. The usefulness of PFS increased from 0.62 to 1.00, which resulted in a decrease in the ICER from $68112.01/QALY to $41417.29/QALY. The ICER values anticipated by sensitivity analysis were, however, less affected by changes in the expenses associated with managing grade 3–4 adverse events (AEs), the tests used, or the hospital fees spent. The ICER was also consistently higher than $37697.00/QALY, as shown by probabilistic sensitivity analysis (1,000 iterations) (Figure 4).

Discussion

This study is the first to examine the cost-effectiveness of modified oxaliplatin and gemcitabine regimens with FU and FA in Chinese patients with unresectable GBC on the basis of findings from a literature search. Adopting the viewpoint of the healthcare system in China. We used the most recent price for first-class hospitals in operation.

To calculate the ICER, we selected the Markov model. Given that PFS and OS are longer in the mGEMOX group, which is consistent with clinical trial data, the overall utility of the mGEMOX group is greater (8). Chinese patients typically have greater utility values than

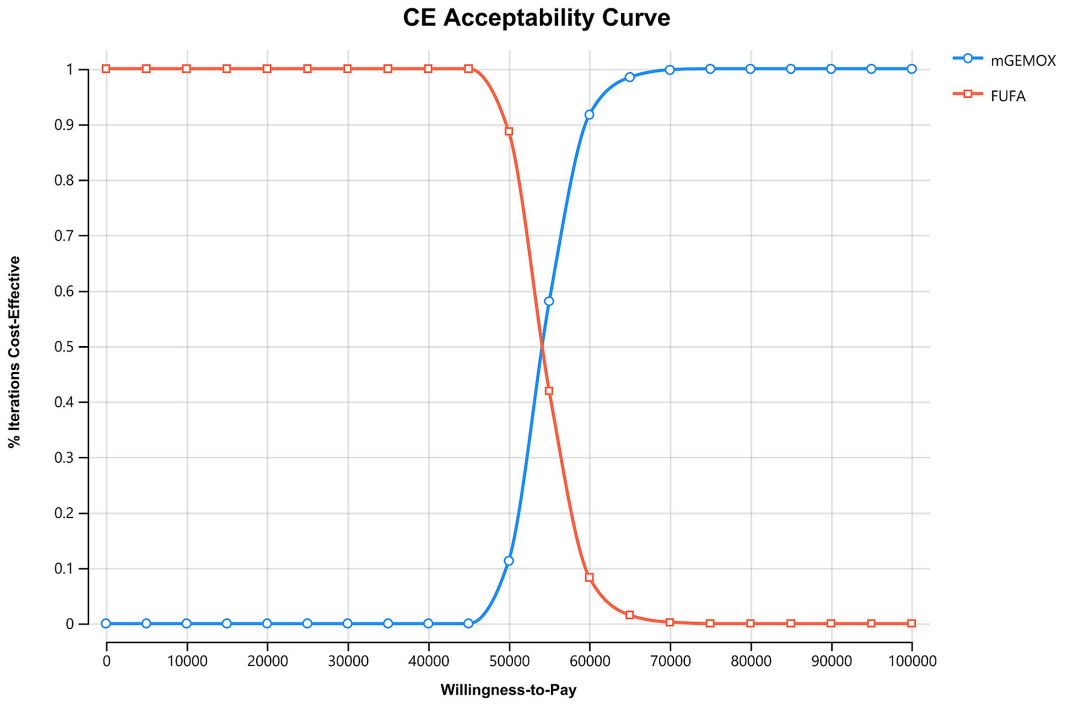

FIGURE 2
Cost-effectiveness acceptability curves. FUFA, fluorouracil and folinic acid; mGEMOX, modified gemcitabine and oxaliplatin.

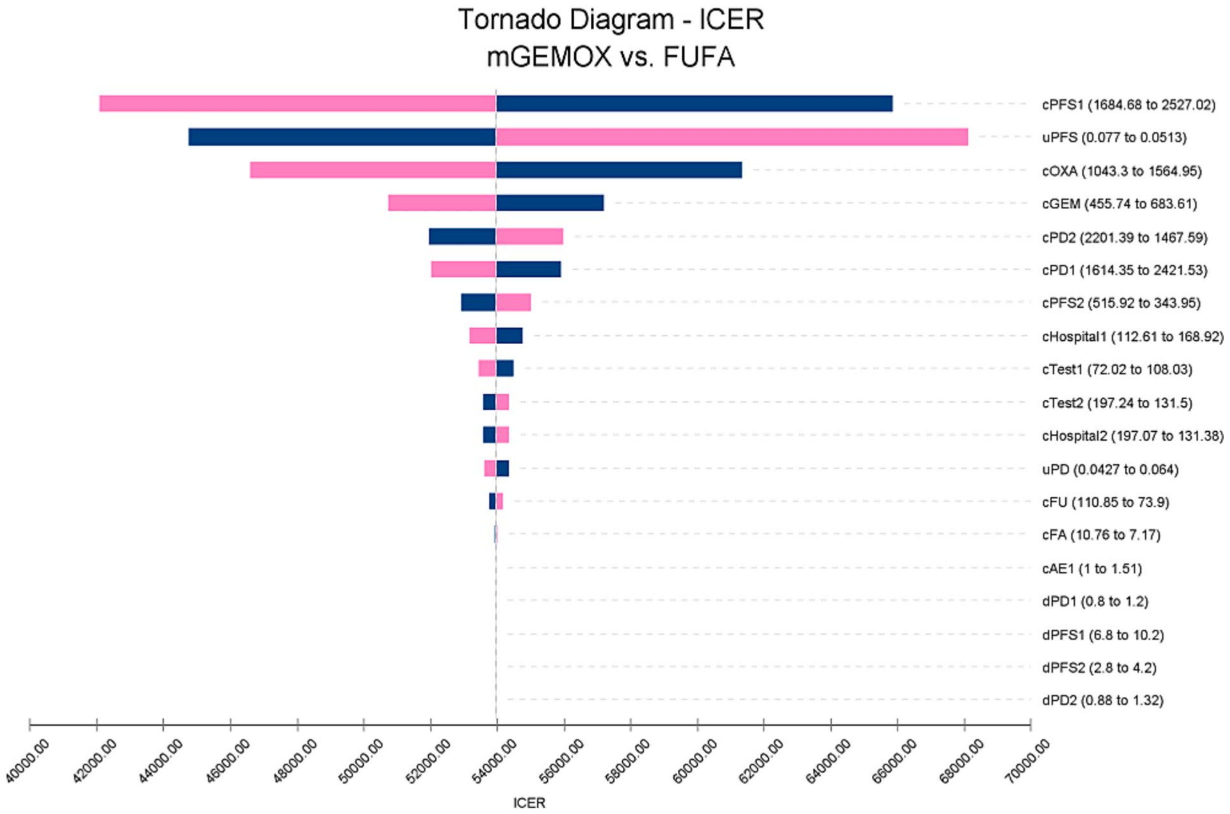

FIGURE 3
Tornado diagram of one-way sensitivity analysis. c, Costs of a specific group; u, Utilities of a specific group; d, Duration of a group; 1, mGEMOX group. 2, FUFA group.

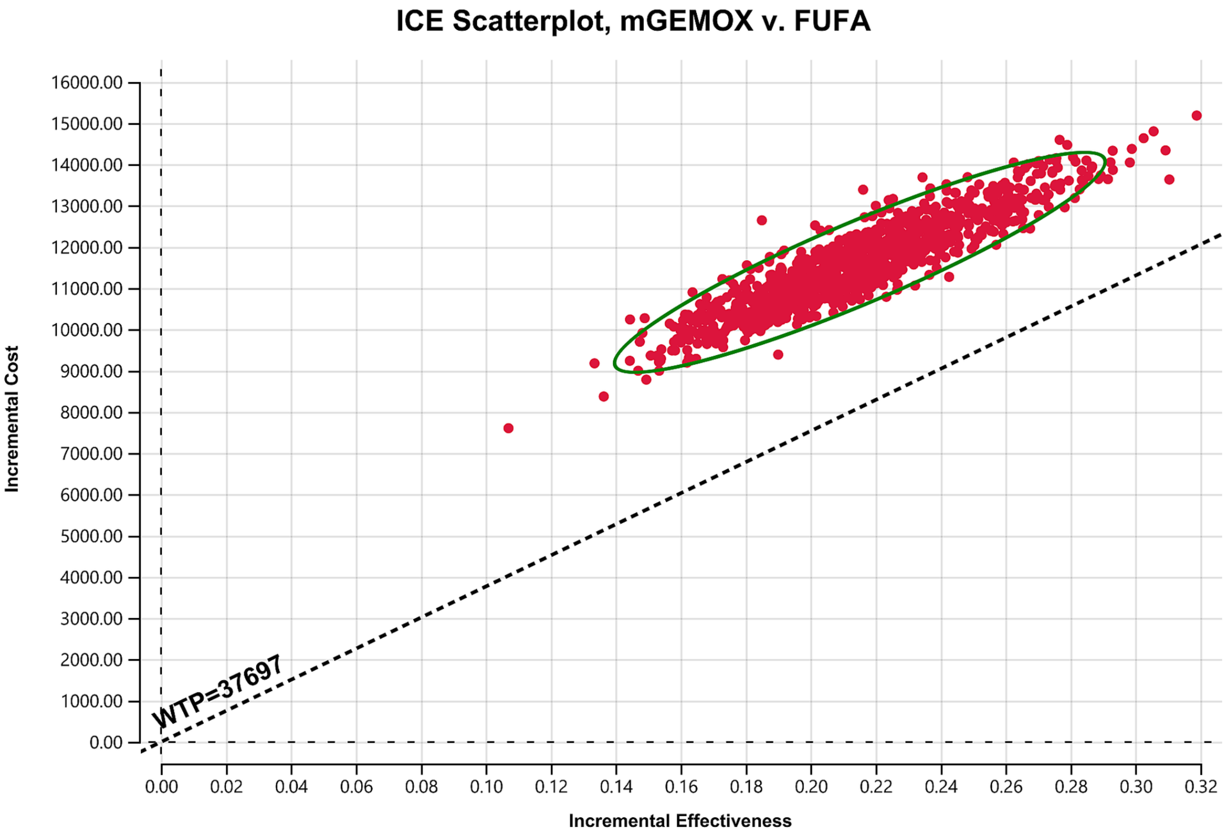

FIGURE 4
Scatter plot of the probabilistic sensitivity analysis results. FUFA, fluorouracil and folinic acid; mGEMOX, modified gemcitabine and oxaliplatin.

do patients in some other industrialized nations because they frequently conceal their genuine ailments, and doctors primarily notify their families (14). China is a vast country with uneven development across different regions. In areas with stronger traditional beliefs, patients tend to rely more on the advice of family members and may conceal some of their symptoms. This contrasts sharply with the more individualistic approach to seeking medical treatment commonly seen in major cities (15). Moreover, China's medical resource allocation also suffers from significant imbalances, primarily manifested in notable disparities between urban and rural areas, among different regions, and across various tiers of medical institutions. This imbalance results in inequalities in the quality and accessibility of medical services, which have profound impacts on residents' health. Policy adjustments and resource optimization are necessary to narrow these gaps and enhance the overall level of medical care (16). This viewpoint is also reflected in the sensitivity analysis. We discovered that although the cost acceptance rate of the mGEMOX group steadily increases when the willingness-to-pay threshold for each QALY is between US$37697.00 and US$52765.59, it is still not cost-effective. The test composition is a cost-effective option only when the WTP value is more than $52765.59/QALY compared to the control group.

The third-generation platinum anticancer drug is oxaliplatin. It is a platinum-based diaminocyclohexane chemical that is effective against ovarian and colorectal cancer. It was introduced in France in 1996, and the FDA gave its approval in 2002. A new cytosine nucleoside derivative called gemcitabine works mostly in the G1/S phase. The NCCN advised gemcitabine and oxaliplatin for biliary cancers because of their survival advantage (17), but there are variations in the regimens' economic reports among nations. In Japan, treating advanced biliary tract cancer with cisplatin, gemcitabine, and gemcitabine is not cost effective (18). A cost-effective therapeutic option for advanced biliary cancer in the US is cisplatin with gemcitabine, which can replace gemcitabine as a single drug (13). By examining the treatment of advanced biliary tract cancer from the standpoint of China's health service system, Chen et al. (19) demonstrated that the capecitabine + oxaliplatin regimen is more cost effective than the gemcitabine + oxaliplatin regimen as a first-line therapy. However, our data demonstrate that the modified gemcitabine + oxaliplatin strategy is not more affordable than the fluorouracil + calcium combination (19). The median PFS for the mGEMOX group was reportedly 8.5 months, whereas it was 3.5 months for the FUFA group. Compared with those of fluorouracil and leucovorin, the utility value of modified oxaliplatin combined with gemcitabine improved by 0.22 QALYs after the model was run for 10 years, but only at the current Chinese exchange rate ($37697.00/QALY). Owing to the increase in overall cost, modified gemcitabine plus oxaliplatin is not an affordable treatment option for unresectable GBC. The cost ratio between the two patient groups with PFS disease status reached a high of 11.4%.

The guidelines suggest the use of oxaliplatin, a member of the third-generation platinum class, as the initial therapy for a number of tumor types. Another representative Phase II study also demonstrated the clinical efficacy of this regimen. This study included 31 patients with previously untreated advanced biliary cancer (19 of whom had gallbladder cancer), all with good performance status and serum bilirubin levels below 2.5 times the upper limit of normal (ULN). When treated with gemcitabine (1,000 mg/m^2, administered on Day 1) + oxaliplatin (100 mg/m^2, administered on Day 2) every 2 weeks, the response rate was 36% and the median overall survival was 14.3 months (20). The cost of medical insurance in China has dramatically decreased in recent years as a result of numerous discussions, although the ICER is still significantly greater than the WTP level. Patients with unresectable GBC now have a chance of survival due to mGEMOX; however, the different treatment methods used in the Chinese medical system are financially hindered by high drug prices and a lack of medical resources. Provinces with high GDP, however, should consider adding oxaliplatin paired with gemcitabine to the local supplemental list, given the encouraging treatment gains reported.

In this study, the Markov decision tree model was utilized to simulate disease progression. However, certain limitations should be noted: the extrapolation is inadequate, and the cost–benefit analysis relies on clinical trial data rather than real-world studies. For the studied population, medical costs were sourced from the Sichuan Province Drug Price Publicity Network and adjacent hospitals. Additionally, the trial data employed in this research originated from local hospitals in India, with no Chinese patients participating in the trials. Consequently, the findings are more suitable for guiding health policy decisions in western China.

In conclusion, the findings of this study demonstrate that, from the perspective of Chinese society, mGEMOX is not economically advantageous for GBC patients compared with FUFA. To make mGEMOX more relevant for this patient population, it should be considered to appropriately reduce costs and offer social aid.

Data availability statement

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

Ethics statement

The data for this study are sourced from publicly available literature and do not include any intervention measures with human participants or animals. The ethics review committee of West China Hospital of Sichuan University granted exemptions for patients' informed consent.

Author contributions

ZC: Writing – original draft, Conceptualization, Investigation. FT: Writing – review & editing, Data curation, Methodology.

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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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EDITED BY
María Del Carmen Valls Martínez,
University of Almeria, Spain

REVIEWED BY
Peter E. J. Baldwin,
Health and Safety Executive, United Kingdom
Ana Sanchez Perez,
University of Almeria, Spain

*CORRESPONDENCE
Christine Clavien
✉ christine.clavien@unige.ch

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Behind the good of digital tools for occupational safety and health: a scoping review of ethical issues surrounding the use of the internet of things

Maéva El Bouchikhi[1], Sophie Weerts[1] and Christine Clavien[2]*

[1]Swiss Graduate School of Public Administration, University of Lausanne, Lausanne, Switzerland,
[2]Institute For Ethics, History, and the Humanities, Faculty of Medicine, University of Geneva, Geneva, Switzerland

Introduction: The internet of things (IoT) is increasingly used for occupational safety and health (OSH) purposes in private and public organisations. Current practices and regulations are unclear, and some stakeholders raised concerns about deploying this technology at work.

Methods: Following the PRISMA-ScR checklist, we reviewed the main opportunities and ethical issues raised by using IoT devices for OSH purposes, as discussed in the academic literature. We searched peer-reviewed papers from 2008 to September 2023, written in English and available in “Web of Science,” “PhilPapers,” and “Google Scholar.” We found 1,495 articles, of which 61 fulfilled the selection criteria. We classified ethical topics discussed in the papers in a coherent description frame.

Results: We obtained 6 overarching categories: “ethical opportunities,” “surveillance and problematic data re-purposing,” “difficulty to inform, consult, and obtain consent from employees,” “unintended and unpredictable adverse effects,” “suboptimal data management,” and “external factors that are conducive to ethical issues.” The resulting list of problematic issues is unexpectedly furnished and substantial. Such a list provides information and guidance for those who wish to develop evaluation frameworks in line with a preventive regulatory approach. It also informs policymakers and practitioners about the governance of such tools for ensuring more OSH.

KEYWORDS

internet of things, ethical issues, occupational safety and health, opportunities, surveillance, adverse effects

1 Introduction

Private and public organisations increasingly deploy connected devices such as sensors, that continuously collect and communicate data. These devices, also called Internet of things (IoT), often involve algorithmic systems and complex AI technology. They can contribute to automating arduous tasks, optimise the use of space, monitor employees’ pace of work, or even reduce the risks of work-related accidents and diseases. They may help to detect early burnout, support healthy lifestyles, promote well-being, facilitate medical treatment or detect disease transmission in an epidemic context (1). They may also take a great variety of forms, such as connected chairs, smart watches for corporate wellness programmes, stress sensors, risky behaviour trackers (e.g., fast driving), safety material (e.g., helmets, boots) with built in GPS, or contact tracing devices (2, 3).

Their contribution to better health and more safety at work allows them to be characterised as tools for fostering the agenda for decent work promoted by international organisations.

However, media, international agencies, non-profit organisations and scholars warn of problems related to the deployment of technological solutions, including IoTs, in the workplace and beyond. For example, in 2016, the Guardian released that Uber experienced a massive data breach after having extensively monitored and collected the personal data of its employees (4). In 2023, a scandal broke out at Queen Mary University in London, because space utilisation tracking tools were deployed without consulting faculty members and students (5). The *European Agency for Safety and Health at Work* highlighted that the adverse effects of hidden surveillance count among the main future challenges posed by IoTs, automation of tasks, artificial intelligence, and autonomous decision-making systems (6–8). Similarly, the *European Digital Rights* (EDRi) and the non-profit-organisation *Worker Info Exchange* (WIE) report evidence that traditional and gig economy workers are more controlled, managed, and subject to algorithmic surveillance through sensors, facial recognition, and other automated-decision-making systems than before (9). All these elements highlight the reinforcement of a surveillance society, exacerbated in the workplace through the deployment of algorithmic management (10, 11).

Yet the risks of using IoT for occupational safety and health (OSH) have remained a blind spot in policy and academic work. At the level of regulation, lawmakers have been sensitive to numerous challenges posed by digital transformation at work while overlooking the risks of IoT deployed for OSH. For example, the European Union has categorised as "high-risk" some types of AI systems deployed for optimising employment, worker management or access to self-employment. Precautionary measures and transparency obligations have recently been enforced in the new regulation, imposing limits on the devices that can be put on the market or used to make decisions on recruitment, promotion, termination of contractual relationships or task allocation based on algorithmic analysis of individual behaviour or personal traits or characteristics (12). Similarly, in the literature, although the challenges posed by the digitalization of the workplace are largely debated, little research is specifically dedicated to the acceptability of IoT for OSH purposes. Relevant issues are addressed, but mostly in passing or not in a structured and targeted way.

We consider it as important to make a sound and full-fledged critical assessment of any information technology before deploying it. For such a task, it is important to foster fundamental knowledge about the ethical pros and cons of deploying IoTs for OSH purposes. For this, we conducted two studies: a scoping review, presented here, and a qualitative research including stakeholders, published separately (36).

This paper offers a comprehensive list of the benefits and ethical issues raised by IoTs for OSH. This fundamental knowledge should provide the necessary understanding for building robust and practical ethical frameworks. Moreover, it should enlighten policymakers regarding any generalisation of digital tools for implementing OSH policy and their evaluation methods. Finally, it should help stakeholders engaged in labour and OSH policies: employers, unions and OSH practitioners.

In the next section, we describe the methods we used to conduct our scoping review. We then present our results, which we classified into benefits and negative impacts. We follow up with a discussion section on the main common trends among this variety of problems. In conclusion, we draw future research avenues.

2 Methods

To conduct our scoping review, we followed the PRISMA-ScR checklist (13). We searched in "Web of Science," "PhilPapers," and "Google Scholar" for articles, proceedings, or editorials, published or accepted in early access, after 2008, the date of the important inaugural international *Internet of Things* conference (14, 15). We choose these three databases because together, they enable the identification of high quality and influential articles produced in the fields of science and technology, philosophy and ethics, and the social sciences.

Since there is no established or standardised term for our topic of investigation, we started with a broad search strategy, in order to identify relevant synonyms and spelling variations on relevant key terms covering the topics of ethics, IoTs and OSH. We notably used "ethic*," "ethics," "internet of things," "IoT," "technology," "artificial intelligence," "big data," "AI," "wearable," "health monitoring," "algorithms," "workplace," "work," "occupational health," "safety," "health." We used these keywords with OR and AND relations and adapted our search queries to the features and content of the databases (see Supplementary File 1 for details). Research and selection criteria were elaborated collaboratively between the authors. The first author conducted the search and selected the articles. The second and third authors double checked the selected articles.

We selected the articles based on the quality of the journal, and on the content of the title, abstract, and a quick key-word search in the main text in case of doubt. For this, we applied the following selection criteria: the article [is published in an English peer-reviewed journal (to ensure minimal academic quality)] *AND* [discusses ethical opportunities and issues of IoT] *AND* [includes an explicit discussion on OSH] *OR* [discusses with some detail at least one case of IoT used for OSH purposes]. Since our research topic is rarely discussed as such in the literature, we had to keep our selection criteria broad enough in order to find relevant articles. Moreover, while reading the selected article, we included additional articles fulfilling our selection criteria based on snowballing research around the most cited or relevant items that we found (16).

We read the selected articles and took note, in a common file, of the opportunities and ethical issues that can relevantly be applied to IoTs for OSH purposes. The first author read all the selected articles, while the second and third authors read a subsection of the material. 50% of the material was read by at least two authors. In a common file, the topics were progressively added by all authors while the third author grouped and described them in overarching categories, composed of subtopics. Categories and subtopics were reorganised until we obtained a logically coherent description frame. This work was done in an interactive process involving all authors. There is no pre-registered protocol for this scoping review.

3 Results

The database search was carried out from August to September 2023. 1,495 articles were found, of which 47 items were accessible and fulfilled our selection criteria (see Supplementary File 1 for details).

TABLE 1 List of selected articles, numbered as they are cited in the result section.

1.	Khakurel et al. (49)	2.	Chan et al. (33)	3.	Ajunwa et al. (20)
4.	Yassaee et al. (79)	5.	Naous and Mettler (2)	5.	Schall et al. (80)
7.	Maltseva (54)	8.	Richardson and Mackinnon (81)	9.	Moore (66)
10.	Lupton (52)	11.	Jacobs et al. (43)	12.	Ajana (19)
13.	Gabriels and Coeckelbergh (38)	14.	Moore (15)	15.	Moore and Piwek (67)
16.	Moore (68)	17.	Bovens et al. (26)	18.	Marinescu et al. (55)
19.	Burr et al. (29)	20.	Mettler and Stepanovic (59)	21.	Mettler and Stepanovic (60)
22.	Brous and Janssen (28)	23.	Baldassarre et al. (22)	24.	Pouyakian (73)
25.	Mejia et al. (62)	26.	Leclercq-Vandelannoitte (51)	27.	Tucker et al. (82)
28.	Costantino et al. (34)	29.	Calvard (30)	30.	Mcaleenan et al. (61)
31.	Molaei et al. (65)	32.	Oravec (83)	33.	Sestino et al. (84)
34.	Tamers et al. (85)	35.	Abioye et al. (18)	36.	Bavaresco et al. (24)
37.	Patel et al. (86)	38.	Svertoka et al. (87)	39.	Suder and Siibak (88)
40.	Pütz et al. (89)	41.	Weerts et al. (90)	42.	Cebulla et al. (31)
43.	Chalaris (32)	44.	Jetha et al. (45)	45.	Segkouli et al. (91)
46.	Sharon (92)	47.	Malomane et al. (53)	48.	Kabir and Alam (46)
49.	Bowen et al. (27)	50.	Pasquale et al. (93)	51.	Howard (41)
52.	Six Dijkstra (94)	53.	Le Feber et al. (50)	54.	Segura et al. (95)
55.	Iavicoli et al. (42)	56.	Martinetti et al. (56)	57.	Nihan (96)
58.	Niknejad et al. (97)	59.	Martinez-Martin et al. (58)	60.	Martinez-Martin et al. (57)
61.	Karale (47)				

Snowballing research added 14 further items to the list, bringing the total to 61 selected articles (see Table 1). We could not access 8 articles that seemed to fulfil our research criteria based on reading their abstract. Paywalls blocked these articles, and their authors did not respond to our request to send us a copy.

During the analysis process, we approached saturation of new relevant topics discussed in the literature after reading 40–45 articles. Here we provide a summary of the main topics identified. We grouped them into 6 overarching categories composed of subtopics and described them with some illustrative references. The resulting description frame is summarised in Table 2.

3.1 Ethical opportunities

IoTs for OSH purposes can generate ethical opportunities for workers, employers, and society. First, these devices may positively contribute to preventing work-related safety risks and health problems. They may diffuse useful and timely information, inputs, and counselling to workers, leading them to become more aware of OSH risks, to gain knowledge on themselves (i.e., early illness detection, identification of weaknesses or bad habits), to increase their individual responsibility over health, to empower them to set and achieve personal health goals, to take more care of their safety, to gain assurance in their relationship with health professionals, and possibly with researchers working with their data. Some IoTs provide the possibility to reach out to others for helpful counselling or debriefing, to communicate and facilitate exchanges with those who share similar hardships, or to exchange experiences. These interactions enforce solidarity, socialisation, and community experience. IoTs may also reduce the burden of physical and unsafe tasks, train and nudge workers to adopt safe and healthy working habits, or help optimise their work environment, an asset for maintaining healthy workers. In specific contexts, besides health and safety, some IoTs may provide employees space for more autonomy or flexibility (e.g., working from home may otherwise not be allowed). From these points of view, IoTs can serve as effective means to decrease fatalities, to improve health prevention, workers' capabilities and productivity at work, work-life balance, and global and well-being and healthy lifestyle (1–49).

These devices are also useful for employers that will be more aware of non-optimal working conditions (e.g., information about stress levels at work or about factors that increase the probability of hazards). Indeed, through the data collection and analysis process, IoTs make it possible to make decisions or formulate recommendations that are more objective, accurate, equitable, and free of prejudices because they are based on a large amount of data collected continuously and analysed in a standardised way, a mark of reliability (7,9,23,35,43,44,46,52,53). Such information will help employers to enhance the quality of risk assessments and diagnosis of OSH situations, simplify the management of risk avoidance measures, optimise emergency responses, facilitate accountability in case of injuries, assess and demonstrate the efficiency of OSH measures (1,4,5,7,11,15,17,20-23,25,28,31,34,35,38,40,41,43-47,49-51). Moreover, IoTs can help employers identify vulnerable groups in specific conditions and tailor subsequent interventions for them (21,23,25,34,49).

TABLE 2 Overview of topics discussed in the literature.

Ethical opportunities	
Surveillance and problematic data-repurposing	Surveillance
	Re-use for dubious purposes
Lack of information, participation, and consent	Lack of participation
	Lack of informed and free consent
	Abuse of trust
Unintended and unpredictable adverse effects	Negative impacts on employees' physical and mental health
	Weakening of workers' power, competencies and self-confidence
	Negative impacts on working processes and habits
	Risks of breaches of sensitive information
	Pressure for normalisation and exacerbation of social stigma
	New or increased existing inequalities
	Weakening of human relationships at work
	Weakening of trust relationships at work
	De-responsibilisation of employers
	Normalisation of surveillance practices
	Dystopian and dehumanised workplaces
	Increased controversies surrounding IoTs and AI in public discourses
	Disruption drivers of public health and employment policies
	Negative environmental impact
Suboptimal data management	More data than needed are collected or stored in the long term
	A too large spectrum of stakeholders has access to the data
	Lack of secure systems
	Low quality of data or of data analysis
	Numerous devices deployed in the workplace
Contextual factors that are conducive to ethical issues	Blurred limits between professional and private life
	Techno-optimism and lack of ethical awareness among stakeholders
	Emergency situations
	Complexity and lack of competencies
	Lack of scientific evidence or peer-reviewed data
	Costs of ethical safeguards
	Lack of clear overall governance

Finally, the deployment of IoTs may have several large-scale societal benefits. If broadly deployed in multiple companies, they can help curb the dissemination of a contagious illness or alert to the outbreak of an epidemic risk. Aggregated information can also contribute to research, with positive downstream effects on public health, clinical practices, and the optimisation of healthcare delivery and OSH practices (10,19,23,34,46). These devices may also contribute to optimise workflows and reduce the costs related to medical care (12,43).

3.2 Surveillance and problematic data-repurposing

Surveillance and information re-purposing is the first set of ethical issues that the deployment of IoTs for OSH purposes can generate.

3.2.1 Surveillance

Authors express strong worries about the hidden surveillance made possible with IoTs and autonomous decision-making systems (9,41). "*The way these technologies are used in organizations has led to the development of new forms of control, which seem more insidious, subtle, and misleading than past forms of IT-based control* (e.g., *computerized performance monitoring*), *in that they are less visible, are indirect, and are often disguised with a rhetoric of emancipation and autonomy*." (26). IoTs allow employers to collect, visualise, and analyse large amounts of personal and sensitive worker data (5,10,21,31,48,54). This massive data collection enables performance analyses and the monitoring of workers and is conducive to surveillance practices that benefit companies, such as excessive managerial control or problematic intrusion in employees' privacy (1-7,9,12-16,18-21,24-26,29-32,46,48,49,51,55-58). For instance, some taxi companies have implemented sensors to detect factors that endanger safe driving (e.g., prolonged working hours, speed, and signs of driver fatigue). These devices help avoid car accidents but can also be used to detect and fire poor drivers (42). In this context, IoTs increase the asymmetric power relationship between employers and workers, especially the most vulnerable (1,3,29,40,52).

Surveillance becomes ubiquitous with IoTs that can also be easily relocated and capable of collecting a wide range of data. Once one form of justifiable surveillance (e.g., a necessary means for sending individualised safety warnings related to a work task) is enforced and accepted, it can be abusively extended beyond its original scope (e.g., for controlling workers' pace of work) (5,7,48). In extreme cases where data become enmeshed in larger networks, by promoting the use of IoTs for OSH purposes, private companies may contribute to some forms of State and international surveillance, as exemplified by contract tracing apps connected to datacentres of national agencies (10,16,29,39,46,48,54).

3.2.2 Re-use for dubious purposes

Even in the case of IoTs primarily deployed for OSH purposes without surveillance goals, once the collected data are at disposal, they still may be *re-used for dubious purposes*. This risk of "function creep" (expansion of the use of a system or technology beyond its original purposes) is high considering the ambiguous interest of companies: they need workers in good health while expecting them to be loyal and productive. In this respect, IoTs provide extensive ways to collect information and monitor workers' behaviour or performance, sometimes without their awareness (3,5,7,12,14-18,20,21,23,24,26,30,39-43,45,46,48,49,51,55). For instance, a manager disposing of a flux of data on individual employees' locations, originally used for contact tracing to combat the COVID-19 pandemic, may be tempted to check an employee's attendance at work or to verify whether one particular employee is telling the truth when claiming compensation for injuries while working. In another example, early indicators of chronic disease revealed by a wellness-promotion device are used for discriminatory firing, task ascription or promotion practices (1,3,7,12,18,20,39,40,53). Data can also be transmitted or bought by third-party companies, or State administration (3,10,12,18,25,27,30,32,39,41,42,52,54,55,59). Some authors also point out the issue of “free labour,” designating unpaid transmission of sensitive information to third parties that benefit from it (4,5,15,46).

However, the dual use or re-purposing of data is not necessarily considered problematic, notably when used for valuable goals such as research or quality control. In such a case, these secondary goals, and the means to achieve them (i.e., who has access to what) must be properly assessed and justified, notably by showing that such type of re-purposing is likely to be generally accepted, including by the first users of the devices, or by showing that the users can benefit from the products or services resulting from the secondary use of their data (30,59).

3.3 Lack of information, participation, and consent

A further group of topics discussed in the literature is related to respect for employees’ points of view. As primary beneficiaries of OSH measures and directly concerned by the surveillance dimension of IoTs, their opinion and consent are of first importance. However, the scholarship shows that such a point is often not undertaken with the required minimum of rigour, care, and honesty. More broadly, three types of problems emerge: a lack of consultation, of informed consent and a risk of abuse of trust.

3.3.1 Lack of co-designing

Employees are often not properly included in co-designing and decision processes ahead of the deployment of the devices. Moreover, fairness issues may arise if a consultation procedure only involves a subsection of representative employees. Such situations fail to respect the value of workers’ point of view and their autonomy (7,9,17,19,21,26,28,30,41,42,48,50,53,59). Workers have insider knowledge about what could be improved and their security needs; their participation in the developmental stage of technology can also help design more efficient tools. If not consulted, workers may be provided with IoTs they do not want and resist using, or with IoTs that fail to target the most important work security issues, or whose default settings cannot be adapted in ways that make the outcome decisions of the system meaningful (2,21,30,42). Moreover, employees may have different sensibilities about data security issues. For instance, they may disagree fundamentally with IoTs collecting sensitive data about them, or they may have context-specific reactions (e.g., allow devices to provide relevant information to health providers or to national health authorities, but not if their employer can access them). This context sensitivity tends to be overlooked in the rush to develop and deploy technical solutions for good OSH purposes (17,25,38,39,58).

Nevertheless, it may ironically happen that employees demand the deployment of the technology and agree to the implicit constraints and risks to which they then will be subjected. In such situations, the ethical issues are co-generated by the victims, and the difficulty is to protect employees against their judgement and decisions (26,58).

3.3.2 Lack of informed and free consent

When employers plan to use IoTs, they are expected to obtain employees’ consent. Nevertheless, *informed* and *free* consent is seldomly obtained. Often, workers tend to be insufficiently informed, or transparency is lacking about what will be collected, from whom, for what aim and for whom’s profit (1,3,12). In other cases, the relevant information is transmitted in an unfriendly or non-understandable way. Users (especially those with little literacy or ease with the technology) are likely to be unfairly overwhelmed by the burden of gathering and understanding all relevant issues, a task requiring skills that cannot reasonably be expected from users (1,10,19,28,29,37-39,41,42,47,54,55,59-61). Workers rely heavily on employers’ willingness to consult them and to provide them with relevant information and choice options. High-quality consent often involves complex procedures and explicit responsibility taken on the part of employers, which they may be reluctant to engage in. Sometimes employers have strong incentives, such as industrial secrecy, for retaining part of the relevant information (4,7,16,17,19,39,52,54,55,59-61).

Furthermore, the line between voluntary and compulsory consent is not always clear-cut. For instance, due to their contractual relationship, power asymmetry, peer pressure, or lack of the privacy prerequisite for an autonomous decision, employees may not be free to refuse or negotiate. If they do so, they may face negative consequences such as being stigmatised or marginalised by employers or peer workers, or excluded from goods or services that rely on the collected data or on the use of the device. In general, it is more difficult to reject practices that are already largely accepted among workers (3,8,13-15,17,29,30,40,41,46,58). Moreover, asking for free informed consent from a population that is vulnerable in many respects (e.g., less educated, from a different social background, with precarious health), and not necessarily aware of the importance of consenting, requires pro-activity and communication skills that employers and occupational health staff seldomly master (10,54,55,59). Consequently, in many situations, the consent obtained is of poor quality (e.g., based on external pressure, biased presentation of information, misleading short-term benefits) and sometimes partly fabricated (3,15,18,25,32,39,41,46,53,56,60). In some cases, deployment of devices or data collection is practised without any form of consent. For instance, this is the case with devices that capture data in private contexts where family members are interacting with the employees. It is also the case with devices that are widely distributed or that operate over long periods of time, one may forget to ask consent. In the latter case, for instance, periodic consent renewal is rarely done, although it is important to remind users of the ongoing monitoring (7,43,61).

In the case of data reused by third parties in aggregated form for external purposes such as medical research, device optimization, or public health goals, consent procedures are even more difficult to follow and often poorly practised. For instance, it remains challenging to provide useful information about possible linkage and recoupling of data by third parties, or about the risks of disclosing sensitive information about specific social groups (46,54,59).

Finally, deployment of technologies that have been insufficiently consented to can elicit practical difficulties such as poor adherence or acceptance of the device from workers. It also raises major ethical issues such as disrespect of workers’ autonomy, and all sorts of sweeping consequences that workers would have resisted if they were informed (5,20,32,40). In a worst-case scenario, if, for some reason, employees fail to use an IoT properly, this could lead to the collection of bad quality data. Unknown to the employees, these data may then be used to train models (deemed to become suboptimal) or to make wrong predictions about employees’ health status. This in turn may lead to suboptimal or problematic managerial decisions. Without proper information and consent procedures, employees are oblivious of the causal role of their original failure and have no way of avoiding such detrimental causal chains (23,24,46).

3.3.3 Abuse of trust

The literature highlights a third type of problem regarding workers: the risk of abuse of trust. In some cases, workers may place unconsidered trust in their employer or in the health specialists within the company and, therefore, may not objectively balance risks and benefits while being consulted about the device. This lack of critical view is a sign of vulnerability which leaves room for possible abuse (3,5,17,49,59). In other cases, workers may not be conscious of the extent of surveillance or dual-use practices because they are not transparently informed and the device is invisible or seems (deceptively) harmless (28,38,56,60,61). The discovery of such abusive secondary uses will, of course, be detrimental to trust relationships. When deploying IoTs, an additional task for companies to fulfil is to help maintain trust relationships with employees (33,53).

3.4 Unintended and unpredictable adverse effects

A third and large set of ethical issues is related to the adverse effects that can occur with the deployment of IoTs for OSH purposes. Their potential autonomous continuous learning processes and prolonged use may generate major structural and cultural changes within organisations and beyond. We identify no less than 14 adverse effects in the literature, and the authors elaborate on an important list of issues related to unintended and (partly) unpredictable short-or long-term adverse effects of deploying IoTs. They highlight that these effects are not given sufficient consideration and often become apparent during late testing phases or after the deployment of the technology, by which time it is too late to make changes.

3.4.1 Negative impacts on employees' physical and mental health

The feeling of being constantly monitored by invasive tracking systems that blur the limits between professional and private life may, in the long run, generate feelings of being under continuous threat, significant fatigue, stress and anxiety. These states of mind are conducive to burnout (4,9,14-16,25,26,34,44,51,56). The deployment of IoTs may also involve additional tasks related to the device or organisational changes that further destabilise employees or generate fear (15,29,33). Moreover, devices developed for creating self-tracking habits can generate cognitive overload, headaches, some forms of cyber-sickness, health-related anxiety, an excessive and damaging obsession with personal health, techno-stress, or addictive self-tracking among users (1,5,13,15,19,24,26,28-30,34,37,38,42-45,50,53,56-58,61). Even if devices may be helpful for reducing some risks, when co-deployed with a demand for an intensification of work, this effect can counterbalance any safety and health benefits (9,16,51).

In case of poor design, IoTs may provide wrong or biased information or directives or generate a limited and poor view of broader social and contextual factors that are important for safety and health. They may be burdensome to use, especially for some groups of workers, such as elder workers. They may be badly secured and create new unintended health or safety workplace hazards, such as mechanical, electrical, thermal, or chemical risks. These issues are particularly problematic when the implemented devices have little or marginal demonstrated health-related benefits, or when they are introduced as a replacement to traditional and more effective OSH methods (2,3,6,17,19,21,28,30,32,34,35,43,44,46,50,51,56,59).

3.4.2 Weakening of workers' power, competencies and self-confidence

Another adverse effect is that workers may progressively lose some competencies (e.g., despecialisation, decrease of critical thinking or risk alertness) because the devices take care of some of their previous duties. In the long run, workers who overly rely on technology for assessing health and security risks may become less skilled at identifying these risks (7,9,21,34). Conversely, users may become unsure about their own perceptions of judgement when the devices produce outputs in contradiction with their evaluation (e.g., about current stress level or about what should be done), or they may not feel entitled to rely on their evaluation when the device fails to spot a risk that they have seen. As pointed out by Mc Aleenan(30) *"there is a potential for a taught helplessness syndrome emerging from this type of technology where workers rely on it to inform them rather than on their own observations and judgement. Conversely if the worker, in the absence of an alert relied on direct observation to stop work, how would this be interpreted by management."* Some authors even argue that prolonged use of IoTs that track and send feedback on a reduced set of personal features may generate a reductionist understanding of health and selfhood and profoundly alienate users from their true selves (1,16,19,29,46). Moreover, the devices may require workers to acquire additional technical skills that they struggle to master (10,18,19,43,51). Overall, the prolonged interaction with the devices may affect not only employees' agency, workplace role, and competencies but also their self-image and self-esteem, and generate feelings of frustration, of being disrespected, of inferiority and subordination to machines, and possibly feelings of incompetence and fear of job loss (13,18,21,26,28,30,31,37,42,44,47,56).

3.4.3 Negative impacts on working processes and habits

An IoT can also be a cause of distraction from work if it generates over-dependency on self-tracking or if it triggers unproductive and disruptive competition between workers (1,13,20,21,24,26,28,29,37,38,42,56). It can alter working practices if employees are aware of being observed or because they are incapable of adapting to new workflows (9,16). Backfiring effects such as reward hacking or new rule-breaking behaviour may occur. For example, employees may develop new behavioural strategies to circumvent the obtrusiveness of an IoT or to neutralise an employer's excessive collection of personal information; thereby they may take more safety risks or increase communication barriers in the workplace (5,6,20,21,44,49). In the opposite direction, the ubiquity of the system can lead workers to set excessive constraints on themselves. For instance, they may be so constantly responsive to the inputs of devices that they work to exhaustion (26). Moreover, when IoTs fulfil tasks that were previously completed by workers, they can unsettle employees and working routines by challenging conventional knowledge and expertise (4,7,14,20,40,42,44,61).

3.4.4 Risks of breaches of sensitive information

Another issue may arise when managers, co-workers, or external third parties (individuals, companies, State) acquire knowledge of private information about workers that was previously hidden (6,21).

Massively collecting and storing sensitive data increases the risks of data theft, data leaks, or unwarranted disclosure of personal information. Information breaches can happen in non-ordinary situations such as cases of cyber-espionage, but also in ordinary working contexts such as when security alerts targeted at one worker are heard or visible to other workers (22,25,43,48,51,53,58). These risks are more likely to occur when workers' data sovereignty (i.e., right to access and to exert control over their personal data) is not guaranteed or made reasonably accessible because workers do not use the device to protect themselves against malicious use, if the devices used are not technically robust against hacking, or if the data transmission and storage process is not managed in a secured manner (3,21,25,40,41,45,59). One can easily lose track of the life cycle (i.e., where and for how long are the data accessible or transmitted to whom) of data that can be copied and stored (45,52,61). In some cases, information breaches can even be a risk for the company itself, such as when strategically relevant internal deficiencies are disclosed to competitive external actors (17).

3.4.5 Pressure for normalization and exacerbation of social stigma

Since IoTs allow for large-scale quantification and comparisons between workers, new expectations or social norms at work oriented towards healthiness and effectiveness may be shaped. The phenomena of co-surveillance, inappropriate competition between workers, or social pressure towards healthy behaviour or towards presenting oneself as being healthy may arise (7,13,30,41). Such trends of normalised expectations at work usually operate at the expense of individuality and individual differences (7,21). This can have negative impacts on workers, especially those who are already fragile or who do not conform to or meet the normative criteria induced by the health-promoting technology. For instance, exaggerated fitness goals generate health risks and social shaming of those who do not meet the standard (8,10,13,16,32,37,41,45,46,49,59). More generally, a large deployment of IoTs may create pressure for conformism and normalised behaviour, with its correlated effects: loss of individual initiative, self-contained or innovative decisions, despecialisation of workers' job duties and activities, decrease in workers' acuity in evaluating OSH risks, and blurring of individual responsibility in case of hazard (1,2,7,28-30,40,42,46,56).

3.4.6 New or increased existing inequalities

Workers may also experience various forms of discrimination, stigmatisation or exclusion (by employers or colleagues, in the workplace or above). It can be the case because IoTs disclosed information about workers' health risks, or due to workers' refusal to use the proposed device, or their difficulty conforming to normalised expectations generated by the devices, or the overtaking of some of their tasks and competencies by the technology, or on their lack of ease with the technology (1,3,7,12,15,18,29,42-46,48,52,57). Regarding the latter issue, the useful notion of "digital divide" and its overlap with existing vulnerabilities is also discussed (34,48,51). In all these instances, some categories of workers will be more at risk of suffering in one way or another because they feel insecure, stigmatised, unequally treated, experience true discrimination, etc. Older generations of workers, workers who are more likely to develop work-related illnesses, or minority ethnic groups will be particularly impacted by the unequal distribution of benefits and hazards of the deployed technology: IoTs may affect differently or under-perform on these groups of workers because they do not fit with the "ideal worker" in light of which the IoT is configured, or because they do not master the technology (due to literacy or language barriers), or because the devices' output reveals "unliked" sociocultural features of workers, creating avenues for a series of unequal treatments (income gaps, unfair work-tasks distribution, etc.) (1,10,15,21,27,29,43,44,48,59). In this context, some authors discuss the notion of "occupational health inequity," referring to "*avoidable differences in work-related fatalities, injuries, and illnesses closely linked with social, economic, and/or environmental disadvantages*" (34). Despite OSH goals, the right to fair working conditions may be insidiously endangered for some categories of workers (19).

3.4.7 Weakening of human relationships at work

If IoTs replace humans in security and health prevention messaging, they will modify social interactions and communication paths, or teamwork in the workplace. Such a situation may generate communication errors or break important relational ties (9,13,18,28,42,56). To illustrate, an automated system may create unforeseen communication issues by providing incentivising messages that are difficult to understand by end-users, thereby generating stress and mistrust among employees (44,45,59). The prolonged use of devices designed to avoid risky human interactions at work (e.g., a contact tracing app during the Covid-19 pandemic) may contribute to social isolation, social anxiety, and hostility among workers (16). Improved safety measures with IoTs that enable an intensification of the workload may also decrease social relationships. In this context, employees may be unequally willing to endorse and able to engage with the technology, and some may feel abandoned or dehumanised (24,28,30,34,37,44,56). Overall, turning some OSH tasks over to IoTs may weaken communication, understanding, and compassion between employees and occupational physicians or health and safety managers. In some cases, workers may even end up creating inappropriate ties with the technology, by attributing intelligence, empathy, or trustfulness to an AI system (42).

3.4.8 Weakening of trust relationships at work

Even when IoTs are deployed for addressing important health and safety issues, the above-mentioned feeling of being constantly monitored, evaluated and possibly discriminated, and the weakening of human relationships may endanger trust relationships between employees and managers, occupational physicians or OHS managers, and weaken safety and health management and culture. Mistrust and a correlated reluctance to use IoTs is most likely in case of abusive surveillance, when the outputs of the devices are based on opaque algorithms, when the deployment of IoTs is not properly explained and accompanied by transparent information about the reasons for the deployment, the associated risks and alleviation measures, or when workers have not provided explicit consent (1,4,5,7,20,30,32,35,38,40-42,45,49,51,53,59).

3.4.9 De-responsibilisation of employers

Considering that IoTs are generally embedded in AI systems that can make assisted or autonomous decisions, part of OSH responsibility may be out-or subcontracted to the machine and thereby shifted outside existing managerial or company protocols. This raises all sorts of understanding and regulating difficulties. For instance, it may blur the channels of internal accountability in case of adverse events or professional illnesses, and lead to a back-scaling of employers' OSH legal obligations (42,44).

Moreover, as they have done something by deploying IoTs for OSH purposes, employers may feel discharged from the duty of implementing further prevention measures or healthcare services (15,16,29). While discussing these issues, some authors point out the risk of over-responsibilising employees for their security at work or for their health status, since they have received OSH counselling and inputs by the devices (8,12,14,31,45,46,53). The increasing deployment of IoTs for health purposes in working and in private contexts is accompanied by sweeping expectations that workers (and citizens) pay an active role in self-tracking their health and safety risks, and in caring for themselves. They thereby become responsible for managing those risks. If they fail in this task despite the support provided by the technology, they tend to be held responsible for the hazards and illnesses that plague them. Such a trend, which is observable not only in working contexts but in society at large, is correlated with a de-responsabilisation of employers and public health authorities, and with a decrease in funding for professional social support and healthcare services (1,10,29,46,59).

3.4.10 Normalisation of surveillance practices

In the long run, surveillance may become the default norm rather than a measure that needs to be justified. Such a change of paradigm may occur gradually without stakeholders noticing. As surveillance practices become more commonly used in a variety of contexts (home office, business travels, etc.), workers may become less aware of the obvious associated risks, especially in cases of passive monitoring (no particular action needed from users) where workers are not regularly made aware of the fact that they are monitored or in the context of gamification practices when individual progresses can be scrutinised by "followers" or compared against others within seemingly harmless challenges or competitions (3,12,15,17,24,29,42,46). This can gradually lead to acceptance of more exacerbated or invasive forms of surveillance (e.g., facial recognition, multiple data collected on the same individual, etc.), possibly up to the "internet of bodies" with devices inserted into humans, and to a society where everything is connected through an "Internet of Everything" (43,46,59). Such forms of intensive monitoring are usually of interest to companies that commercialise or deploy the devices at the expense of workers (32,56,59).

3.4.11 Dystopian and dehumanized workplaces

IoTs are also portrayed as potentially contributing to the emergence of dystopian scenarios where a great range of decision-making becomes automated and where employees are submitted to excessive algorithmic management, meaning that the workload attribution, time management, or short-term on-demand employment of vulnerable workers is managed by automated AI systems. A large deployment of IoTs may blur the boundaries between AI and humans, raising the question whether standards originally designed for ordinary human workers still hold (15,18,42,44,51). The risks of dehumanising the workplace and its negative repercussions are abundantly discussed by some authors (7,15,18,42,44,56). Workers may lose their individuality, autonomy, expertise and value within the company and become opportunistically utilised as "*farmed and domesticated entities (..) rather than autonomously involved in authentic health maintenance initiatives*" (32). If workers have little choice but to accept workplace monitoring despite its constraining and dehumanising effect, it may stifle their motivation and creativity while fostering suspicious beliefs (14,30).

3.4.12 Increased controversies surrounding IoTs and AI in public discourses

As we have seen, IoTs for OSH purposes may contribute to the general trend of workplace surveillance and generate numerous adverse effects. Workers and citizens are likely to become aware of these ethical issues, as is the case when media disclose how targeted advertisements stem from massive repurposing of data collected by IoTs. Such awareness often does not facilitate public discourse on tracking devices, and on AI in general. It can generate strong controversies conducive to a polarised society (19,32). Some authors regret the lack of transparent information about flaws in the technology and in its underlying algorithms, arguing that it is an obstruction to productive public discussions on how AI systems can be improved. Mistrust towards technology and AI is also problematic if it hinders the deployment of technologies that permit socially valuable advancements (18,35,44).

3.4.13 Disruption drivers of public health, social values, and employment policies

In the long run, the widespread deployment of IoTs for OSH purposes may also have a wider impact on the social understanding of health and safety issues. IoTs enable efficient assessments, predictions, and behavioural recommendations without imposing solutions. Stakeholders are mostly responsible for following (or not) the automated injunctions made by the devices. In this sense, extensive use of such technology reinforces the neoliberal understanding of social healthcare duties: the domain of OSH tends to be reduced to prediction and information tasks, leaving employees the responsibility of taking care of their health capital. The rhetoric of users' empowerment is put forward while promoting IoTs, leaving in the shadows the employers' and state's duties to provide social healthcare to ill workers and citizens (2,3,8,9,12,15,16,29,46).

In parallel, health data philanthropy is promoted as a means to promote medicine and healthcare. Related to this trend, one author worries about a correlated depreciation of the value of privacy, depicted as individualistic and as an opposite of openness, transparency, and public good. Employees are increasingly expected to contribute to the common good by taking care of their health and by giving their private and sensitive data (12).

A further issue is the gradual increase in importance of private health industry. OSH is one place among others in which private companies developing health-related devices colonialise the public health domain, thereby disrupting traditional public tasks and responsibilities (7).

Similarly, IoTs for OSH purposes contribute to the enforcement of neo-Taylorism practices in the workplace. Employees are exposed to daily automated injunctions are quantified and assessed in multiple ways by AI systems and are sometimes favoured (or disfavoured) based on those assessments. Such work organisation compromises workers' individuality and capacity to organise and form resistance to these new management tools and contributes to the social divide between those who have power and those who are reduced to quantified workers (1,14,16,21). These practices may have a widespread impact at the societal and policy level, notably by weakening the social aspect of employment policies (1,14,16,29,43,46,51,55).

3.4.14 Negative environmental impact

Last identified adverse effect is the significant environmental impact (carbon footprint, environmental consequences of the extraction of rare material, recycling issues) of such connected technology, which becomes a serious issue in time of global climate change and environmental insecurity (9,14,31,56).

3.5 Suboptimal data management

We identified another group of five ethical issues in the scholarship. They concern suboptimal data acquisition, transfer, storage, processing, and dissemination. They are linked to situations already mentioned and are particularly likely to occur in the case of IoTs that collect sensitive health information and may be deployed at workers' homes in the context of a home office.

3.5.1 More data than needed are collected or stored in the long term

First, there is the temptation with IoTs to collect more data than needed for OSH purposes. For instance, additional data may be collected for the purpose of personalising the outputs of the systems. Sometimes, additional data are collected without clear goals, just because the default setting of the system allows it, or just in case these data could be relevant later. Collecting a wide variety of data on the same individuals during a continuous period of time in different contexts (work and home) are factors that increase the risks of deanonymisation of sensitive data at the expense of equal treatment. Problematic situations also occur when data are collected at a time for a particular purpose, but stored longer than needed, thereby increasing the risk of later reuse. This was an issue at the end of the COVID-19 crisis (39). Once a worker's profile has been created in a database, it can "persist" endlessly. These are all cases of breach of the principle of data minimization and are conducive to numerous ethical issues such as excessive surveillance, increased mistrust in workplaces, etc. (5,12,18-20,32,38,39,42,49,52,59-61).

3.5.2 A too-large spectrum of stakeholders has access to the data

Second, depending on the device's purpose, various actors may have access to the data, such as the IT unit and services, managers, co-workers, OSH staff, third-party medical institutions, network infrastructure suppliers, government bodies, etc. It may be unclear (or insufficiently managed and controlled) where to set a limit on who is authorised to access what data, for what purpose and until when. For instance, it is often unclear why human resources should receive health data collected for OSH purposes (12,17,25,45,49,51,53-55,59). In numerous situations, third-party companies are also given the task of storing or processing the data (e.g., conducting the data analysis). These companies may insidiously resell part of the data (e.g., to private health companies interested in the health profile of future clients) or use them for other purposes (e.g., for sending targeted ads to users) (1,13,41,53,56,59).

3.5.3 Lack of secure systems and procedures

Next, IoTs may not be robust enough to react against unauthorised data access by third parties, cyber-attacks, or malicious uses, either due to lack of inbuilt security measures, poor conception, or to failed maintenance. The collected data may be insufficiently de-anonymised, allowing breaches in personal privacy when re-identification (usually with correlative analysis on different datasets) is operated by a third party (1,3,5,12,17,18,22-24,27,28,31,35,37,38,42,43,48,54-56,58,60,61).

3.5.4 Low quality of data or of data analysis

Moreover, imprecise or partly irrelevant data, as well as the use of suboptimal or biased algorithms, may lead to unnuanced or incorrect interpretations. For instance, this may happen if the data used for training the system are not representative of the target OSH domain in the workplace or if the technical system is unable to represent the diversity of workers and their characteristics in an appropriate format. When those limitations are overlooked, inappropriate or discriminatory outputs and decisions may occur or be reinforced (2,3,7,9,14,15,17-19,23,25,32,34,35,41,42,44,46,48,49,51,52,55,59).

3.5.5 Numerous devices deployed in the workplace

Finally, if an expanding number of devices is deployed, each with its own purpose, technical specificity and database, it becomes difficult to ensure a centralised governance and effective security controls (39,61). It may be a further cause of distress for employees who do not feel at ease with the trend towards technological advancement (18).

3.6 Contextual factors that are conducive to ethical issues

Contextual factors compose the last major category of topics discussed in the literature. Even though they are not ethical issues in themselves, specific contextual factors may be conducive to ethical issues.

3.6.1 Blurred limits between professional and private life

If used in the context of a home office or on portable devices such as smartwatches, IoTs contribute to blur the limits between professional and private life, especially when surveillance practices extend over strict working activities and when they operate in the background, more or less outside user awareness. This reality induces organisational difficulties (e.g., how to set clear boundaries between working and free time) or increased stress levels among workers who may feel constantly monitored in their daily routine and struggle to take breaks from work. It may also generate a lack of clarity about employers' rights and duties (4,5,7-9,12,17,21,24,26,34,41,42,45,46,55-57).

3.6.2 Techno-optimism and lack of ethical awareness among stakeholders

Stakeholders may lack critical thinking. Developers of IoT and AI systems are not always clear about the spectrum of possible wrong uses or problematic motives for using their products. Employers deploying the solution, may be overenthusiastic and readily assume that the technology is objective and efficient, that it will benefit the workforce, without foreseeing its limitations or being aware of the risks of lack of efficacy, data re-purposing by third parties involved, etc. For instance, employers readily accept methodologically limited whitepapers produced by the companies that commercialise the IoTs, and if at all, the efficacy of the devices is usually examined after their commercialisation and deployment in the workplace [15,19,21,22,24,46,51].

If relevant stakeholders show such overconfidence in technological solutions, combined with low ethical awareness, one can expect little vigilance and motivation to measure appropriate outcomes, no systematic risk assessments of all process stages, and a lack of mitigation measures. Stakeholders may be satisfied with superficial checks for legal acceptability. They may systematically underestimate foreseeable risks and associated harms and overlook the ambivalence of their use of the technology when it implies some form of surveillance [26,30,42,46,59]. For instance, when IoTs are used to assess employees' health conditions, only a narrow range of easily measurable or quantifiable criteria can be taken into account, leaving out relevant contextual factors such as individual or socio-cultural characteristics. If such limitation is not taken seriously due to an over-optimistic appreciation of the device, biased or suboptimal OSH measures may be taken by OSH staff and occupational physicians. Consider the further illustration of an IoT provided to cleaning ladies in the hotel industry to assist them in case of sexual assault. In that case, location tracking capabilities of the device need to be particularly developed to quickly find the worker and send assistance: efficacy is correlated to a high degree of surveillance, and employers are likely to undermine one or the other difficulty [17,19,25,55].

Overall, human-centred policies and ethics-by-design procedures have not yet become standard or harmonised. This absence can amplify the adverse impact that IoTs have on employees. Authors also worry that stakeholders may have to deal with too rigid and fixed ethical frameworks, which make them ill-equipped to deal with the novel demands of a rapidly evolving technology [19,41,42,44,51,53].

3.6.3 Emergency situations

Emergency situations are a contextual factor that can bear up the deployment of IoTs. One known example is the contact tracing apps during the COVID-19 crisis, which were strongly endorsed by employers to reduce the propagation of the virus. In such a situation, there is less time, funding, and mental availability to evaluate the appropriateness of the purposes and to consider the long-term adverse effects of a technology. Once it is deployed, there is a risk of keeping the solution in the long run without a proper re-evaluation procedure or despite the counterevidence of its proportionality [38,39,45].

3.6.4 Complexity and lack of competencies

The fast-evolving field of technology means that the developer's community may lack consensus on how to use, integrate or optimise the technology and at the end of the chain, users may be insufficiently informed [1,37,43-45,56]. A myriad of devices are developed at a fast pace and deployed for a large variety of applications. From the development to their implementation, numerous actors, decisions, and information transfers are involved. Each can be limited in many ways. For instance, some IoTs allow for automated decision procedures based on systems containing self-learning algorithms. Consequently, they raise explainability issues: even designers of the devices do not have an operational understanding of how the devices make predictions or recommendations[22,48,51,60]. Some systems are grounded on strong theoretical assumptions that are not clearly spelled out or justified: this is typically the case for devices supposed to measure well-being, a state that can be defined in many ways and tracked with a large variety of observable features[19]. Operational conflicts may occur at the deployment stage of the IoT: since developers do not use common languages, operating systems, and evaluation practices, compatibility issues with other technologies used in the same workplace are likely to emerge[37].

During the implementation stage, actors involved (medical staff, OSH team, workers, public health decision-makers) may lack proper training and education [9,22,24,29,43,55]. Moreover, once data are collected, given the number of stakeholders involved, it remains particularly challenging to keep a reasonable control on the flow and use of data [19,61]. Overall, when new IoTs are deployed for the first time, it is difficult to foresee all possible hazards. To some extent, it is a form of unregulated social experimentation [14,22,26,28,42,43].

The complexity of the technology and data process makes it particularly difficult to provide clear and comprehensive information allowing stakeholders to anticipate how workers may be impacted by the deployed IoT. It becomes more challenging to respect workers' rights over their data [9,24,28,48]. This is particularly true when workers lack the digital literacy and competency to use and understand the technology, or when the devices are regularly updated, iteratively developed, redesigned and extended, or when they include opaque machine-learning algorithms that cannot be traced or controlled by humans [10,28,30,37,42,43,45,52].

Such complexity generates new illiteracy and its correlated risks. The difficulties involved in understanding, explaining, and evaluating IoTs and AI systems and their outputs are obstacles to stakeholders' (especially workers') trust in the deployed technology and their capacity to manage it. Some stakeholders may not even try to gain minimal technical knowledge, which does not help informed decision-making [1,2,22,24,35,43,47,52,53].

3.6.5 Lack of scientific evidence or peer-reviewed data

In many cases, there is a lack of high-quality scientific evidence (e.g., based on randomised control tests and study designs with high external validity) that the devices are valuable tools for achieving their goals (taking into account long-term efficacy and adverse effects) in the particular workplaces, compared to more human solutions. This makes it difficult to evaluate whether it is proportionate to implement them in the first place or to keep using them once deployed [2,3,6,25,32,35,37,41,42,44,45,51,53]. For instance, not enough (or not clear enough) measures are collected, leaving too much space for interpretation, and making it difficult to evaluate whether a device positively contributes to work health or safety [2,56]. Interestingly, the difficulty of accessing high-quality data is sometimes caused by legitimate privacy protection measures (data anonymization, limited access to sensitive data) set up to prevent opportunities for abuse [1,40]. Or it may be due to market constraints: companies are reluctant to disclose the parameters of their algorithms and to provide insight into the data used to train their AI systems because private investments are at stake [44].

A connected topic is the difficulty of evaluating the accuracy of the outputs produced by theory-based algorithms trained on selected datasets that are not always close to the workplace context or to the typology of workers who will use them. Deep-learning systems are even more difficult to evaluate since they work in a non-transparent manner [1,3,37,58,59].

3.6.6 Costs of ethical safeguards

The deployment of IoTs that fulfil ethical standards often involves huge additional time and costs: investment in the development of more secure systems (devices, software, etc.) which require more technological expertise; development of more complex implementation strategies that take care of workers' vulnerabilities; organisation of proper information and training for managers and users; ensuring long-term maintenance and security monitoring, etc. It is unclear who is willing and who should pay for these costs (developer companies or companies who buy the devices), especially in the context of a competitive market. Strong conflicts of interest are at stake: for instance, when monetary incentives are involved, it is difficult to produce an objective cost–benefit analysis of security measures. Moreover, if by some mechanism, strong security measures are imposed on companies (for justified ethical reasons), unfair market advantages may arise, since the costs (especially high when dealing with AI or interconnected systems) may not be affordable for the public sector or small-to-medium-size companies in contrast to big and wealthy firms (1,2,29,31,35,38,40,43,47,50,55).

3.6.7 Lack of clear overall governance

Worries are expressed regarding the lack of specific regulation framing the deployment of such systems, at national and international levels, and regarding the fact that existing legal frameworks that may be applied vary greatly by country. Notably, authors worry about how one should apply data privacy regulation (GDPR), medical device regulation, labour law or contract law to the deployment of IoTs, while taking into account all country-and region-specific challenges (9,12,15,23,24,35,39,41-44,48,54,59,60). Authors also worry about the extent to which companies and third parties that have some access to sensitive data comply with regulations and enforce robust privacy practices (41,42,48,53). A meaningful application of existing laws is particularly challenging due to the complexity of the interconnected technology and insufficient legal and technical literacy among policy and company leaders (1,9,44,60).

The lack of standards, guidelines, institutional safeguards, and regulatory bodies is also an issue. Rights and ethical principles are sometimes too abstract and not amenable. International and national laws are maturing at a slow pace compared to technological innovations. Even when clear and applicable laws and guidelines exist, they remain ineffective if they are not sufficiently well communicated and enforced by regulatory procedures and institutional bodies (3,9,12,22,25,42,44,45,48,51,59). Within the private sector, it is also difficult to find appropriate institutional procedures (e.g., validation processes, regular staff training) or departments that oversee and regulate the use of IoTs by conducting controls and audits, and by delivering authorizations based on strong security controls. There is also a lack of specific protections for vulnerable groups of workers, such as older populations. Consequently, those who develop or deploy the technology in the private sector may not comply with law because of ignorance (or absence) of guidelines and policies, lack of common standards (exemplar methods and practices), absence of strategic frameworks for the development, acquisition and use of IoTs, and overall absence of regulatory control (1,12,19,29,37,42-45,48,50,55,59,60).

Without clear and standardised ethical guidelines, laws, independent regulatory bodies or related agencies, it remains unclear what is expected from developers, deployers, and users of the technology. For instance, to what minimal standards are they expected to abide? To what extent are they expected to engage with contextual trade-offs and interests, such as protecting the privacy of specific vulnerable groups at some additional cost or at the expense of organisational efficacy (15,17,59). It is also unclear who is accountable at each step of the process in case of security or health hazards due to malfunction, error, lack of integrity, or in case of unequal treatment of workers (9,17,18,28,31,37-39,42-44,51,56,60). One illustration is the fact that gig economy workers are less protected by law due to the lack of clarity on their working status (34,59). Another illustration is the case of IoTs deployed with insufficient procedures to handle cases of conflicting evaluations prompted by the device and made by employees, making it difficult to define employees' rights (to contest the systems' outputs) and responsibilities (42,44,48). Without clear regulation and governance, it will also be difficult for workers to know what are their rights (e.g., do they have property over their health data? who else has it?), how these rights are respected by the company (e.g., who has access and possibly owns their data?), whether their rights are protected by the administration, and how they can fight for them (1,3,5,12,32,37,38,45,48,55,56,61). While discussing these issues, some authors regret the current neoliberal political trend tending to less regulation and more individual responsibility on the shoulders of employees (29).

Lack of guidance or constraints enables the flourishing of a competitive market of self-tracking health devices, with products selected for their economic viability rather than ethical acceptability. These difficulties increase if the health care system is not clearly structured and too many companies or individuals can claim some right of access to workers' health data, or in the case of strong business interests (e.g., needs for data collection in a competitive market), or if the pace of the regulatory process is not adapted to the dynamic and short life cycles of technology deployment (29,32,44,49,55).

Effective regulation, standards, and governance is made difficult due to a number of reasons: the inherent difficulty of conflicting rights and constraints (e.g., right of workers' privacy and information, right of industrial secrecy, necessity of business productivity, etc.), the lack of human oversight on what is deployed, the technical complexity of the technology and the difficulty to ensure the interconnectivity between the different systems (e.g., large number of protocols and devices that are deployed across different application domains that are more or less interoperable with new and old systems, and that generate various types of data), the lack of expertise among stakeholders, and the lack of clear task and responsibility attribution to those stakeholders (2,15,24,26,31,42,43,48,55).

4 Limitations

Since very few articles that we found explicitly focused on ethical issues *of IoTs for OSH purposes*, we often had to apply relevant general theoretical points made by authors to our study case or adapt points originally made with a different focus. However, we do not think that by doing so we distorted the original authors' points of view. In most cases, the ethical concerns the authors raised apply to a variety of cases, including IoTs for OSH purposes.

Due to paywall constraints, we did not have access to 8 potentially interesting articles. However, since we approached saturation of content after reading 40–45, it is unlikely that we missed important contributions.

This review does not include grey literature in which additional ethical issues may be discussed. For instance, one interesting highlight that came up during the review process of this publication is the ironic safety risks posed by IoTs for OSH purposes: these devices often involve AI solutions that are limited regarding their explainability, analysability, and relevance of output data. "Black boxes" and limited datasets may generate safety risks or poor health & security standards. Safety monitoring of IoTs for OSH purposes is therefore required (44, 69).

Since the topics presented here overlap and intersect in myriad ways, they could have been organised differently. This methodological difficulty is unavoidable and has already been highlighted by others (21).

5 Discussion

The list of ethical issues resulting from this literature review is impressively (and unexpectedly) furnished, varied, and substantial. Many issues would deserve an extensive discussion that we cannot make in this paper. Some of them echo well-known topics which have also been highlighted in broad-scoping theoretical works [e.g., (21, 39, 48, 63, 64, 70–72)]. Nevertheless, none of these studies focussed on the impacts of IoTs primarily deployed for good purposes such as OSH. This indicates that, these devices are generally not understood to be ethically problematic since they are deployed for noble goals (the health and security of employees) and fit within the new trend of the 5.0 industry that places the well-being of workers at the centre of production processes (73). As a result, they tend to escape the necessary ethical scrutiny (51). The blind spot in the literature and among policymakers is appealing at a time of narrative in favour of technology (e.g., AI for good, data for good). To a certain extent, it also illustrates the framing power of academic and political discourses.

Moreover, among the five categories of issues that we point out, three are focused on direct and tangible elements: the employer, the worker, and his or her data. They refer to easily observable and abundantly discussed difficulties (e.g., lack of consent, surveillance, exploitation, data security) which we expected ahead of conducting the review. We were however more surprised by the two further categories that cover adverse effects and contextual factors. These categories are surprisingly furnished and reveal a deeper layer of complex and interconnected ethical issues that were more seldomly addressed. Our scoping review method helped to reveal these topics discussed in different disciplines (sociology, business ethics, AI ethics) that do not systematically communicate with each other. For instance, some authors in the social sciences literature have already made calls for societal impact assessments, but their concerns did not bridge to other academic fields. In this context, we believe that a robust cross-disciplinary ethical analysis can play a role in better informing IT developers and OSH policymakers and practitioners.

Facing the wide spectrum of ethical issues, and in particular the imposing number of possible unintended adverse effects posed by IoTs for OSH purposes, one should seriously ask the question whether the deployment of these devices should be encouraged or refrained. For such an assessment, the principle of proportionality is very important theoretical tool. It may not be easy to apply it in the case of IoTs because the positive and negative or unintended impacts to be compared in the proportionality evaluation are often of a different nature (economic costs and benefits, physical or psychological effects, levels of trust, etc.) and are also unevenly distributed between the different types of stakeholders (employers, workers, third parties). Therefore, one should be aware that large margins of interpretation remain possible. A second difficulty is the fact that numerous and interconnected effects are at stake and are often difficult to disentangle. Overall, however, we feel that a fair application of the proportionality principle makes it difficult to take a positive view of an IoT deployment for OHS purposes. At minima, it highlights the urgent need for nuanced assessments before deploying such technologies in complex situations.

Our results need also to be read in comparison with the results of a connected empirical study that we have conducted (36). In that study, we used focus groups and individual interview to obtain first hand stakeholder points of view. Our results reveal further ethical highlights that were not explicitly or only marginally discussed in the academic literature. Notably, numerous stakeholders that we interviewed doubted that (in most cases) IoTs could efficiently replace classical OSH tools and procedures. This worry comes ahead of discussing proportionality issues. A linked issue is the difficulty of setting up efficient safeguards (to avoid risks of excessive surveillance, data security, etc.) without compromising the adequacy of the devices. Interviewees were also attentive to issues specifically attached to the roles of people within a company. They worried, for instance, about the competencies and integrity of human resources services while handling data produced by IoTs for OSH purposes. They elaborated on the conflicts of interest of the staff involved: on the one hand, occupational physicians or OSH specialists need to support employees and IoT may help them in this task, on the other hand, they obtain data that are valuable (e.g., for managerial purposes) to their employer. Interviewees also discussed the uncomfortable situation in which direct managers can be placed when they must deploy ethically questionable IoTs within their working unit. Further, they wondered about employers' (existing of new) obligations towards workers when IoTs for OSH purposes are deployed.

Furthermore, our data show the field's dynamic and emphasises the need to situate expressed ethical concerns in a time and political perspective (40, 44). Academic discussions on the implications of 4.0 and 5.0 technology have lately become abundant, as we can see by the articles' publication dates. Even though we used 2008 as the starting date for the review, nearly 90% of the articles we found were published in the last 5 years. One observable trend in this growing literature is scholars' interest in the topic of data, privacy and access to own individual information. This selective ethical attention is probably a result of the adoption of the European Data protection regulation (37). In contrast, other topics are overlooked or only mentioned briefly, although they would deserve more development. This is the case with the emerging environmental impact discussion, which is likely to become a major theme in the coming years in most domains of technology (1, 35). Another important topic, overlooked in the IoT literature but highlighted in discussions about AI in medicine, is the suboptimal continuous assessment and maintenance plans of the technology: outdated devices pose all sorts of risks to users (74, 75).

The dissonance between our research results and the ongoing development of ethical awareness also highlights the limits of the classical preventive approach based on risk analysis, which dominates the regulation of information technology. Scholars have already highlighted the limits of such an approach and the difficulty of anticipating what is unknown (25). Our study shows that short and sometimes automatized checklists of risks are insufficient regulatory solutions for the deployment of AI technology, and in particular for assessing IoTs for OSH purposes. Stakeholders need to stay in charge and should be able to use critical thinking before making decisions about the deployment of such technology.

6 Conclusion

This paper shows that deploying of IoTs for OSH generates an unexpected and substantial list of ethical issues. Moreover, the analysis reveals two under-investigated categories of ethical issues, which cover the impact of adverse effects and contextual factors. In this regard, the paper enlightens decision-makers, occupational health professionals, and end-users (employees) who are unlikely to be aware of all relevant issues. Overall lack of ethical awareness is confirmed by our complementary qualitative study (36), in which we found that relevant stakeholders fail to spot numerous and important issues discussed in the academic literature. Such blind spots are even noticeable in the academic realm since several articles that we reviewed heavily focused on the positive aspects of the technology. None of the more critical articles that we reviewed spotted all the issues listed here. Therefore, this paper offers an analytical grid of five categories that could serve as a source of information for the selection and improvement of existing ethical frameworks [e.g., (10, 17, 23, 40, 63, 74, 76–78)], as teaching material for the training of OSH professionals and occupational physicians, as a springboard for targeted scientific research on the use of IoTs in the area of occupational medicine, and as a call to improving laws and regulations.

This paper paves the way for further research. First, it calls for conducting in-depth studies on the ethical issues briefly listed in the result section. These analyses could be connected to related discussions about AI in medicine. Overall, we think that more critical research should be conducted on the way of elaborating and conducting ethical assessments of evolving technologies. Regulators develop guidelines and frameworks for fostering the deployment of AI systems, but they should give greater consideration to the societal and contextual impact of technology, even of technology originally deployed for good purposes. To conclude, this study's main message is that much more can go wrong than an intelligent and careful stakeholder or academic expert might expect at first glance.

Data availability statement

The original contributions presented in the study are included in the article and in the Supplementary File 1, further inquiries can be directed to the corresponding author.

Author contributions

MB: Conceptualization, Formal analysis, Investigation, Methodology, Writing – original draft, Writing – review & editing. SW: Conceptualization, Formal analysis, Funding acquisition, Methodology, Supervision, Validation, Writing – review & editing. CC: Conceptualization, Formal analysis, Funding acquisition, Methodology, Supervision, Validation, Data curation, 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.2024.1468646/full#supplementary-material

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EDITED BY
Hubert Amu,
University of Health and Allied Sciences, Ghana

REVIEWED BY
Patricia M. Alt,
Towson University, United States
Xinpei Wang,
The University of Chicago, United States

*CORRESPONDENCE
Yan Zhang
✉ yanzhangtangdu@163.com

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Exploring the association between socioeconomic inequalities in chronic respiratory disease and all-cause mortality in China: findings from the China Health and Retirement Longitudinal Study

Zhuo Zhang[1], Guoshuai Shi[2], Faguang Jin[3] and Yan Zhang[3]*

[1]School of Health Services Management, Xi'an Medical University, Xi'an, China, [2]School of Public Health, Xi'an Medical University, Xi'an, China, [3]Department of Respiratory and Critical Care Medicine, Tangdu Hospital, Air Force Military Medical University, Xian, China

Objective: Research on the inequality of chronic respiratory disease (CRD) is limited, and the association between CRD and all-cause mortality is not well-established. Investigating the distribution of CRD and its associated mortality risks is essential for improving CRD conditions and developing targeted intervention measures. This study aimed to explore the relationship between inequalities in CRD and all-cause mortality in China.

Methods: This study utilized nationally representative baseline data from the China Health and Retirement Longitudinal Study (CHARLS, 2011–2020, wave 1–wave 5), including a total of 14,743 subjects. The concentration index was employed to measure socioeconomic-related inequality in CRD, and the concentration index decomposition method was used to describe its influencing factors. Cox proportional hazards regression model was employed to examine the association between CRD and all-cause mortality.

Results: The prevalence of CRD was 11.79% (95% CI: 10.98, 12.66) in China. The concentration index for CRD was −0.050 (95% CI: −0.075, −0.026), indicating a certain degree of inequality in its prevalence. Chronic lung disease (concentration index = −0.046, 95% CI: −0.073, −0.019), asthma (concentration index = −0.102, 95% CI: −0.148, −0.056), and asthma-chronic obstructive pulmonary disease overlap syndrome (concentration index = −0.114, 95% CI: −0.173, −0.055) also exhibited a pro-poor distribution. The decomposition analysis of the concentration index for CRD revealed that age, education level, and economic status played substantial roles in contributing to the observed inequality. Additionally, Cox regression analysis showed that participants with CRD had an increased risk of all-cause mortality (HR = 1.49, 95% CI: 1.34, 1.65).

Conclusion: Inequalities exists in CRDs in China, with the prevalence of these diseases primarily concentrated among economically disadvantaged groups. Additionally, CRD increases the risk of all-cause mortality. Addressing the root causes of economic inequalities and enhancing the educational attainment of individuals with low socioeconomic status can help improve the situation.

KEYWORDS

inequality, chronic respiratory disease, asthma, chronic obstructive pulmonary disease, all-cause mortality

Introduction

Chronic respiratory disease (CRD), primarily chronic lung diseases, are a significant category of non-communicable diseases that pose a serious threat to health (1). Globally, the prevalence of CRD stands at 7.1%, affecting an estimated 545 million people. Ranked as the third leading cause of death worldwide, following cardiovascular disease and cancer, this group of diseases is responsible for approximately 4 million deaths (2, 3). In China, CRD are also widespread, securing the fourth position among causes of death in 2019 and accounting for 10.6% of total deaths in the country (4). Beyond the threat to life, CRD can result in diminished physical function, leading to disability and increased medical costs, imposing a substantial burden on families and society (5, 6).

Asthma and chronic obstructive pulmonary disease (COPD) are prevalent CRD (7). Asthma is characterized by chronic airway inflammation, leading to recurrent wheezing, shortness of breath, coughing, and chest tightness (8). Chronic obstructive pulmonary disease, encompassing chronic bronchitis and emphysema, is characterized by persistent airflow restriction and corresponding respiratory symptoms (9). Global prevalence data indicate that asthma affects approximately 334 million people (10), while COPD affects over 200 million people, with about 65 million experiencing moderate or severe disease, making it the third leading cause of death worldwide. Notably, more than three-quarters of individuals with COPD reside in low- and middle-income countries (11, 12). Asthma and COPD also impose a significant health burden in China. Data from the China Lung Health Study reveal that the prevalence of asthma in individuals aged 20 and above in China is 4.2%, with approximately 45.7 million patients (13). As for COPD, the prevalence is 13.7% in individuals over 40 years of age, affecting nearly 100 million people nationwide (14). Studies have demonstrated the existence of asthma-chronic obstructive pulmonary disease overlap syndrome (ACOS), although there is no consistent agreement on the diagnostic criteria for this condition (15). Moreover, the clinical characteristics of this syndrome are complex, resulting in worse health status, increased treatment difficulty, and a significantly elevated risk of poor patient prognosis (16). Therefore, in-depth research and more effective management strategies for CRD are essential to reduce their burden.

Socioeconomic status serves as a comprehensive indicator of an individual's economic and social standing, used to gauge their social status. Generally, a higher socioeconomic status tends to be positively correlated with better health. Numerous studies have identified an association between socioeconomic status and the prevalence of chronic diseases (17). One study highlighted socioeconomic inequalities among patients with various fatal chronic diseases (18). Moreover, studies conducted on the Slovenian population with chronic diseases have revealed a significantly higher incidence of chronic conditions among individuals with lower socioeconomic and employment status (19). In measuring health inequities resulting from socioeconomic factors, the concentration index is widely employed to assess health equity, and its reliability has been well validated in previous studies (20). Additionally, previous studies have shown that CRD may affect all-cause mortality, and a meta-analysis of cohort studies indicated that patients with asthma had an increased risk of all-cause mortality (21). A large national cohort study also found that patients with COPD had a significantly higher risk of all-cause death compared to those without COPD (22).

Numerous studies have highlighted a robust association between socioeconomic status and CRD. However, different studies have presented conflicting results (18, 23). Additionally, despite previous research analyzing the relationship between CRD and all-cause mortality, the literature on the relationship between CRD inequality and all-cause mortality in low- and middle-income countries, especially in China, remains limited. Furthermore, even within large cohorts, results are not entirely consistent, indicating the need for further research. To fill this gap, we used data from the China Health and Retirement Longitudinal Study (CHARLS) to analyze the relationship between CRD and all-cause mortality among Chinese adults and explore the inequitable status of CRD.

Materials and methods

Data sources and study population

The study utilized 2011–2012 baseline data (wave 1) and follow-up data (wave2-wave5) from the CHARLS. Initiated by the National School of Development at Peking University, CHARLS conducted a national baseline survey in 2011, followed by four subsequent visits in 2013, 2015, 2018, and 2020. This study adopted a multi-stage stratified probability proportional scale sampling method to sample residents over 45 years old in 150 counties and 450 communities/villages across 28 provinces (autonomous regions and municipalities directly under the Central Government) in China. The survey encompassed two main components: a household questionnaire and a physical examination survey designed to collect detailed information from the respondents. The purpose of data collection was to provide fundamental insights into China's aging population, supporting the formulation of more effective policy programs aimed at improving the living conditions of the older adult population. A scientific sampling method was employed to ensure that the large sample of older adult people was nationally representative. The information on chronic lung diseases and asthma was obtained through a questionnaire survey, and any missing samples were deleted to ensure the data's integrity and accuracy. The CHARLS project received ethics approval from the Peking University Ethical Review Committee (IRB00001052-11015), and subjects provided written informed consent before the investigation commenced, ensuring the ethical compliance of the study. Moreover, the present analysis received approval from Xi'an Medical University Medical Ethics Review Committee (XYLS2023077).

In this study, individuals with missing CRD information (n = 278), missing economic status information (n = 293), those under 45 years of age (n = 489), and those lost to follow-up (n = 1,905) were excluded. Finally, 14,743 participants were included in the analysis.

Study variables

Chronic respiratory diseases were the main variable in this study, encompassing chronic lung disease and asthma, as obtained from CHALRS questionnaires. The assessment of chronic lung disease involved the question, 'Have you been diagnosed with chronic lung diseases, such as chronic bronchitis, emphysema (excluding tumors, or cancer) by a doctor?' Asthma was determined through the question, 'Have you been diagnosed with asthma by a doctor?' ACOS (asthma-chronic obstructive pulmonary disease overlap syndrome) was not specifically investigated in this study. Since COPD is a significant component of chronic lung disease (24), and there is no uniform definition for ACOS, the comorbidities of chronic lung disease and asthma investigated in this study were considered as ACOS.

The primary outcome of the study was all-cause mortality. Mortality information was collected from the wave 2 to wave 5, with only wave 2 providing an exact date of death. If participants survived during the follow-up period, their survival time was the interval between the two surveys. If they died, the survival time was the interval from the date of wave 1 to the date of death of the participant, or from the date of wave 1 to the median time of the wave with recorded death.

Covariance

The covariates included in this study encompassed basic information such as age (45–59, 60–74, ≥75), gender (male, female), educational level (illiterate, primary school, secondary/high school, university or above), and marriage status (never married, married, others). Lifestyle factors, including smoking (no, yes), drinking (never, occasionally, regularly), BMI (0–23.9, 24–27.9, ≥28), basic health insurance (no, yes), and preventative health service utilization (no, yes), were also considered. BMI was calculated using the weight/height squared formula, and alcohol consumption was categorized as never, occasionally (defined as less than one drink per month), and regularly (defined as more than one drink per month).

Statistical analysis

First, the concentration index was utilized to assess the inequality of CRD, chronic lung disease, asthma, and the ACOS. The economic status indicator employed was the IM_PCE from the "Constructed Expenditure, Income, and Wealth Database" released by CHARLS in 2017. Economic status was categorized into five groups based on quartiles. To further validate the results, we conducted a stratified analysis of the inequality of the CRD based on age and sex. Data were extrapolated by applying the CHARLS sampling weight (ind_weight_ad2) to estimate the prevalence of CRD among Chinese adults aged 45 years and older. Secondly, we described the basic characteristics of the study population. Continuous data conforming to normal distribution were presented as means and standard deviations, while frequencies and percentages were used for categorical data. Thirdly, Concentration index decomposition was employed to describe the contribution of each influencing factor to the inequity. Finally, a Cox proportional hazards regression model was employed to examine the association between CRD and all-cause mortality. Additionally, a stratified analysis was performed according to sociodemographic characteristics. Stata 16.0 was used for data collation and analysis, with $p < 0.05$ considered statistically significant.

Results

Concentration index

The prevalence of CRD, chronic lung disease, asthma, and ACOS were 11.79% (95% CI: 10.98, 12.66), 10.42% (95% CI: 9.65, 11.24), 3.72% (95% CI: 3.29, 4.20), and 2.34% (95% CI: 2.03, 2.71), respectively. The concentration indexes for CRD, chronic lung disease, asthma, and ACOS were −0.050 (95% CI: −0.075, −0.026), −0.046 (95% CI: −0.073, −0.019), −0.102 (95% CI: −0.148, −0.056), and −0.114 (95% CI: −0.173, −0.055) (Figure 1; Supplementary Table S1). The results indicated a certain degree of inequality in the prevalence of CRD, chronic lung disease, asthma, and ACOS, with the prevalence of these diseases mainly concentrated in the population with poor economic status.

Stratified analysis

To further demonstrate the inequity in the prevalence of CRD in different populations, we conducted a subgroup analysis. We found that several diseases were more prevalent among individuals with lower socioeconomic status in both men and participants aged <60, consistent with the results of the main study. However, ACOS did not show significant inequities in women. No inequities were observed in several diseases among participants older than 60 years (Table 1).

Sociodemographic characteristics of the subjects

A total of 14,743 subjects were enrolled in this study, with an average age of 59.72 ± 9.83, comprising 7,232 males (49.05%). The majority of participants had an education level of high school and below (98.22%), and 86.96% were married. Additionally, 32.33% were smokers, 41.96% were drinkers, and 31.68% were overweight or obese. Moreover, 91.06% had basic medical insurance, and 19.85% had utilized basic public health services in the previous month (Table 2).

Concentration index decomposition

The decomposition analysis of the concentration index for CRD revealed that age, education level, and economic status played substantial roles in contributing to the inequality observed in these diseases. Older age, lower educational attainment, and poorer economic status were identified as factors that elevate the inequality associated with CRD. Additionally, the study found that other factors

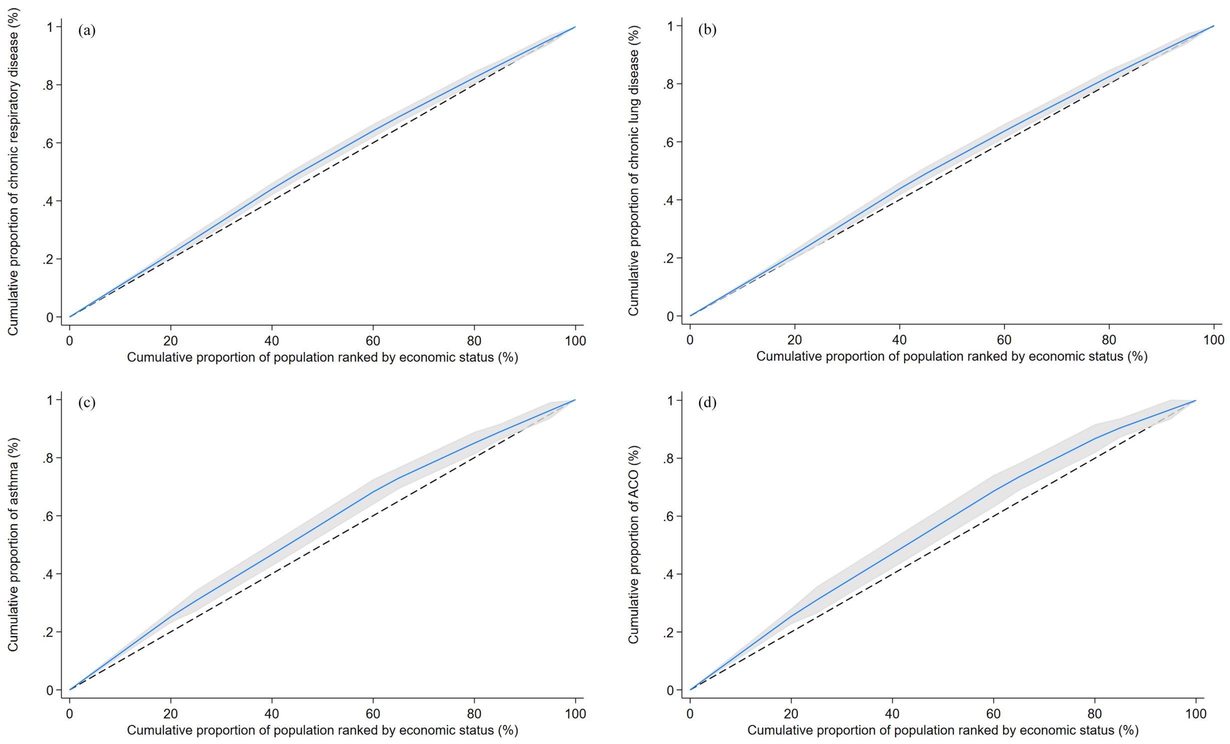

FIGURE 1
Concentration curves for chronic respiratory disease **(A)**, chronic lung disease **(B)**, asthma **(C)** and ACOS **(D)**. The dotted line surrounding the curve represents the 95% CI. ACOS, asthma-chronic obstructive pulmonary disease overlap syndrome.

such as marital status, smoking, and higher BMI also contributed to an increase in inequality related to CRD (Table 3).

Association of chronic respiratory disease with risks of all-cause mortality

Over the approximately 10-years follow-up period, 2,368 deaths were reported, resulting in a mortality rate of 15.76% (95% CI: 15.14, 16.41). Subjects with CRD had a higher all-cause mortality rate than those without CRD (Log-rank test, $p < 0.05$) (Figure 2). After adjusting for all covariates, Cox regression analysis revealed that participants with CRD had an increased risk of all-cause mortality (HR = 1.49, 95% CI: 1.34, 1.65) (Supplementary Table S2). The analysis of specific types of CRD revealed that chronic lung disease, asthma and ACOS were significantly associated with an increased risk of all-cause mortality (Supplementary Table S2). Additionally, stratified analyses based on sociodemographic characteristics showed that while the results for each subgroup varied slightly from the overall findings, there was a consistent trend indicating that CRD was associated with a higher risk of all-cause mortality across different groups (Supplementary Figure S1).

Discussion

We used nationally representative data from the CHARLS baseline survey to examine the socioeconomic inequalities associated with CRD. Our study identified inequalities in CRD and its specific types (chronic lung disease, asthma, ACOS), predominantly favoring individuals of lower economic status. Age, education level and economic status were identified as significant factors contributing to these inequalities. Additionally, our findings indicated that CRD was associated with an increased risk of all-cause mortality. These results highlight the current inequitable burden of CRD and its potential implications for mortality in China.

Previous studies had revealed the inequity present in common chronic diseases, such as hypertension and diabetes (25). This study aligned with those findings, indicating a similar inequality in CRD. Adam et al., utilizing multiple rounds of cross-sectional data from the United States, demonstrated the existence and worsening of socioeconomic inequality in lung health over time (26). Furthermore, socioeconomic status emerged as a potential independent determinant of lung health. On a global scale, inequalities in CRD were also apparent. Haifeng Li et al., drawing on data from the global disease burden, identified a concentration of disability-adjusted life years associated with COPD in less developed countries (27). The socioeconomic status-driven inequality in CRD may be linked to insufficient healthcare programs and inadequate tobacco consumption control (27, 28). These findings underscored the importance of health system reform, emphasizing the need to direct more attention to vulnerable groups based on economic and social status. Additionally, CRD served as predictors of all-cause mortality (29, 30), suggesting that the income-based gap in CRD identified in our study may contribute to life expectancy inequality in China.

This study highlighted the significant impact of age, education, and economic status on chronic respiratory inequality. It was widely recognized that older adults often encounter challenges such as

TABLE 1 Concentration index for chronic respiratory disease, chronic lung disease, asthma and ACOS by gender and age.

	Concentration index	95% CI	*p*
Male			
Chronic respiratory disease	−0.046	−0.079, −0.013	0.006
Chronic lung disease	−0.048	−0.083, −0.013	0.008
Asthma	−0.122	−0.183, −0.061	<0.001
ACOS	−0.168	−0.244, −0.093	<0.001
Female			
Chronic respiratory disease	−0.058	−0.096, −0.020	0.003
Chronic lung disease	−0.045	−0.087, −0.004	0.031
Asthma	−0.078	−0.148, −0.007	0.031
ACOS	−0.033	−0.127, 0.061	0.489
<60, years			
Chronic respiratory disease	−0.073	−0.113, −0.033	<0.001
Chronic lung disease	−0.076	−0.118, −0.033	0.001
Asthma	−0.093	−0.171, −0.015	0.020
ACOS	−0.120	−0.222, −0.019	0.020
≥60, years			
Chronic respiratory disease	0.005	−0.026, 0.036	0.758
Chronic lung disease	0.013	−0.021, 0.047	0.462
Asthma	−0.058	−0.115, 0.001	0.046
ACOS	−0.062	−0.135, 0.010	0.089

ACOS, asthma-chronic obstructive pulmonary disease overlap syndrome.

physical decline and a weakened immune system, increasing their susceptibility to CRD (31). A potential remedy could involve providing increased access to free basic public health services and allocating more resources to medicare drugs related to respiratory health (32). In terms of age and education, economic status emerged as a key modifiable risk factor. Previous research had identified low economic status as a risk factor for CRD (27, 28), exposing individuals to additional risk factors like poor dietary habits and increased occupational dust exposure (33). Conversely, individuals with higher economic status were more likely to leverage medical technology for health maintenance, receive health advice, actively engage in disease screenings (25, 26), and ultimately reduce the risk of CRD.

TABLE 2 Sociodemographic characteristics of subjects aged 45 years and older.

Variables	Categories	*N* (%)
Age group, (years)	45–59	8,289 (56.22)
	60–74	5,185 (35.17)
	≥75	1,269 (8.61)
Gender	Male	7,232 (49.05)
	Female	7,511 (50.95)
Educational level	Illiterate	4,272 (28.98)
	Primary school	5,874 (39.84)
	Secondary/high school	4,334 (29.40)
	University or above	263 (1.78)
Marriage status	Never married	140 (0.95)
	Married	12,820 (86.96)
	Others	1783 (12.09)
Smoking	No	9,976 (67.67)
	Yes	4,767 (32.33)
Drinking	Never	8,553 (58.05)
	Occasionally	1,514 (10.28)
	Regularly	4,667 (31.68)
BMI (kg/m^2)	0–23.9	10,073 (68.32)
	24–27.9	3,351 (22.73)
	≥28	1,319 (8.95)
Basic health insurance	No	1,318 (8.94)
	Yes	13,425 (91.06)
Preventative health service	No	11,816 (80.15)
	Yes	2,927 (19.85)
Economic status	0–20%	3,127 (21.21)
	20–40%	3,077 (20.87)
	40–60%	3,006 (20.39)
	60–80%	2,908 (19.72)
	80–100%	2,625 (17.81)

BMI, body mass index.

Respiratory diseases are a major cause of death worldwide and rank as the third leading cause of death globally (29). In 2017, CRDs were responsible for nearly 4 million deaths, marking an 18 percent increase since 1990 (2). Population-based studies have also reported that CRD increases the risk of all-cause mortality. Bin Zhang et al. summarized the findings of 30 cohort studies, revealing that patients with asthma had a higher all-cause mortality rate (RR = 1.38, 95% CI: 1.07, 1.77) (21). A meta-analysis pooling the results of 12 studies found that even mild COPD was associated with increased all-cause mortality (34). Additionally, a study using data from the National Health and Nutrition Examination Survey found that patients with ACO had a higher risk of all-cause death compared to individuals with asthma only, COPD only, non-asthma/COPD, or non-ACO (35). These findings are consistent with our study. The increased risk of

TABLE 3 Decomposition analysis on the inequality of chronic respiratory diseases.

Variables	Categories	Elasticity	Concentration index	Contribution	Contribution rate (%)
Age group, (years)	60–74	1.633	−0.077	−0.126	37.68
	≥75	0.551	−0.142	−0.078	
Gender	Female	−1.567	−0.004	0.007	−1.28
Educational level	Illiterate	1.604	−0.188	−0.302	45.25
	Primary school	2.025	−0.041	−0.082	
	Secondary/high school	0.695	0.200	0.139	
Marriage status	Never married	0.012	−0.174	−0.002	1.71
	Others	0.156	−0.046	−0.007	
Smoking	Yes	0.226	−0.014	−0.003	0.58
Drinking	No	−0.398	−0.007	0.003	−0.50
	Sometimes	−0.041	0.005	0.000	
BMI (kg/m^2)	24–27.9	−0.279	0.059	−0.016	3.00
	≥28	0.003	0.063	0	
Basic health insurance	Yes	−1.386	−0.008	0.011	−2.11
Preventative health service	Yes	1.09	0.014	0.015	−2.79
Economic status	0–20%	0.01	−0.788	−0.008	18.47
	20–40%	0.213	−0.367	−0.078	
	40–60%	0.06	0.046	0.003	
	60–80%	−0.037	0.447	−0.016	

BMI, body mass index.

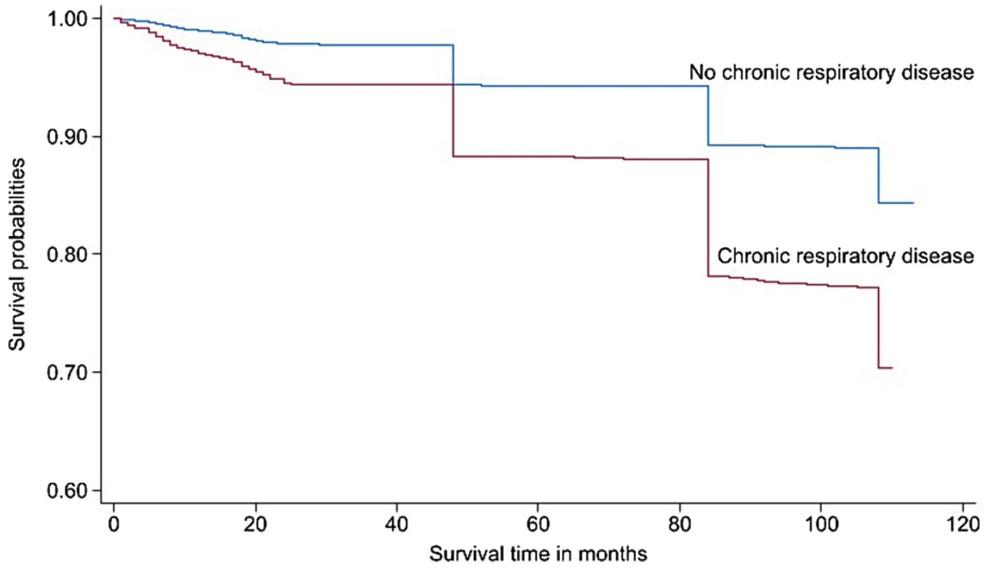

FIGURE 2
Kaplan–Meier curve showing the cumulative risk of all-cause mortality by chronic respiratory disease status.

all-cause mortality in patients with CRD not only from the disease itself but also from chronic inflammation, airway limitation, and comorbidities due to medication (e.g., osteoporosis, pneumonia, depression) (36, 37). Implementing appropriate management strategies and interventions is crucial to reducing this risk.

This study analyzed the inequality of CRD and its risk of all-cause mortality using nationally representative data, providing a crucial foundation for the control of such diseases. However, the article has several limitations. Firstly, the study did not categorize the collection of chronic lung diseases, making it challenging to determine the percentage of COPD. Consequently, chronic lung diseases were used to represent COPD, introducing a certain degree of bias. Nevertheless, considering that COPD is the primary chronic lung disease, this bias is likely minimal. Secondly, the prevalence of CRD relies on self-reporting, which may introduce bias, given the low awareness rates of diseases such as asthma and COPD in China (38, 39). Additionally, due to age limitations in the survey, we were unable to estimate the inequality of CRD in individuals over 75 years of age. Finally, this study employs data from 2011, potentially introducing bias compared to the current reality of CRD inequality.

Conclusion

There is inequality in CRD in China, primarily affecting economically disadvantaged groups, and CRD increased the risk of all-cause death. Age, education level, and economic factors significantly contribute to this inequality. Policymakers and researchers should prioritize the needs of individuals with low socioeconomic status when developing strategies to prevent CRD. Addressing the root causes of income inequality and improving educational attainment among these individuals can help mitigate the issue.

Data availability statement

Publicly available datasets were analyzed in this study. This data can be found at: http://charls.pku.edu.cn/.

Ethics statement

The studies involving humans were approved by The CHARLS project received ethics approval from the Peking University Ethical Review Committee (IRB00001052-11015), and the present analysis received approval from Xi'an Medical University Medical Ethics Review Committee (XYLS2023077). The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

ZZ: Data curation, Formal analysis, Funding acquisition, Methodology, Visualization, Writing – original draft. GS: Data curation, Visualization, Writing – original draft, Writing – review & editing. FJ: Methodology, Writing – review & editing. YZ: Conceptualization, Project administration, Writing – review & editing.

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Conflict of interest

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

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1472074/full#supplementary-material

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EDITED BY
Hubert Amu,
University of Health and Allied Sciences,
Ghana

REVIEWED BY
Juliana Aggrey,
University of Ghana, Ghana
Theodora Yayra Brinsley,
University of Health and Allied Sciences,
Ghana

*CORRESPONDENCE
Ipek Cicekli
✉ ipek.cicekli@acibadem.edu.tr

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High prevalence and co-occurrence of modifiable risk factors for non-communicable diseases among university students: a cross-sectional study

Ipek Cicekli[1,2]* and Serap Gokce Eskin[3,4]

[1]Faculty of Health Sciences, Department of Nutrition and Dietetics, Acibadem University, Istanbul, Türkiye, [2]Institute of Health Sciences, Department of Nutrition and Dietetics, Acibadem University, Istanbul, Türkiye, [3]Faculty of Nursing, Department of Medical Nursing, Aydin Adnan Menderes University, Aydin, Türkiye, [4]Institute of Health Sciences, Department of Medical Nursing, Aydin Adnan Menderes University, Aydin, Türkiye

Background: Non-communicable diseases (NCDs) are a major global concern. This study aimed to examine the prevalence and co-occurrence of lifestyle risk factors among university students.

Methods: This analytical, cross-sectional study was conducted between January and April 2022. A total of 485 students were included in the study. Lifestyle risk variables for NCDs included physical inactivity, low fruit and vegetable consumption, alcohol use, tobacco smoking, being overweight/obese, and sugar sweetened beverages consumption.

Results: Our results showed that the most frequent risk factor was insufficient physical activity (89.2%), followed by low vegetable (70.5%) and fruit consumption (58.9%). Overall, more than half of the students (51.2%) had four or more risk factors highlighting the urgent need for preventive interventions. The co-occurrence of four or more lifestyle risk factors was significantly greater in students at private universities (aOR: 2.01 95% CI: 1.2; 3.35), those living in student homes (aOR: 3.57 95% CI: 1.96; 6.5), and those with fast food preferences when eating outside (a0R: 2.53 95% CI: 1.62; 3.96).

Conclusion: Targeted university-based interventions, such as promoting physical activity, providing affordable nutritious meals, and educating students on healthy lifestyles, are essential to reduce non-communicable disease (NCD) risk among students. Early action fosters lifelong healthy habits, supports healthy aging, and reduces healthcare costs. Future research should focus on refining these strategies to maximize their impact on university populations.

KEYWORDS

noncommunicable diseases, primary prevention, risk factors, students, diet, sedentary behavior

1 Introduction

Non-communicable diseases (NCDs) are a major cause of mortality worldwide with their prevalence rising steadily across all age groups (1). Conditions such as cardiovascular disease and diabetes are increasingly prevalent, driven by modifiable risk factors including poor diet quality, tobacco use, excessive alcohol consumption and insufficient physical activity. To combat NCD-attributable deaths, policies that focus reducing these risk factors and promoting healthy behaviors are essential (2). Behavioral risk factors for NCDs are commonly established during adolescence or early adulthood (3). Behaviors established during young adulthood can persist into later life, increasing the likelihood of developing chronic conditions such as cardiovascular diseases, diabetes, and certain cancers. Moreover, the co-occurrence of multiple risk factors can exacerbate health outcomes, leading to a greater burden of disease (4).

Among young adults, particularly university students, the prevalence of these risk factors is notably high. Studies indicate significant levels of clustering risk factors in this demographic (5). For instance, a study across 24 countries found that 15.9% of university students exhibited three or more behavioral NCD risk factors. Among these, inadequate fruit and vegetable intake was reported by 80.5% of students while 23.1% were classified as physically inactive (6). Another study investigating behavioral risk factors for cardiovascular diseases—such as being overweight, avoidance of dietary fat, low physical activity, tobacco use and excessive drinking—among university students in nine Association of Southeast Asian Nations (ASEAN) countries reported a high prevalence of these risk factors, emphasizing the need for targeted interventions by university health centers and health promotion programs (7).

University students especially vulnerable to NCD risk factors. Research conducted in Bangladesh revealed that over half of the students have obesity (50.4%), with notable prevalence of hyperglycemia (13.5%), and hypertension (12.0%). These findings underscore the critical need for targeted health interventions aimed at lifestyle modifications within this population (8). Early interventions, such as educational programs promoting physical activity and balanced nutrition, could play a pivotal role in preventing the progression of NCDs and reducing the long-term health burden (9).

The impact of NCDs is profound, affecting individuals, families, and national healthcare systems. Direct costs, including medical treatments and hospitalizations, place significant strain on health systems. In the United States and Germany, NCD-related expenditures account for 45 and 51% of healthcare costs, respectively (10). Indirect costs, such as lost productivity, transportation for healthcare access, and employer burdens due to absenteeism, further exacerbate the economic toll. Without preventive measures, these costs will continue to climb (10).

This study aims to assess the prevalence and co-occurrence of lifestyle-related risk factors for NCDs, including poor nutrition, physical inactivity, tobacco smoking, alcohol use, sugar-sweetened beverage (SSB) consumption, and overweight/obesity. The research focuses on students enrolled in the Faculty of Health Sciences at one public university and two private universities, providing insights into the health behaviors of young adults in higher education settings. By identifying these patterns, this study seeks to inform effective public health strategies and interventions that address the rising burden of NCDs.

2 Methods

This analytical cross-sectional study was conducted over 3 months (January–April 2022) at the Faculty of Health Sciences in two universities—one public and one private. Students were approached in their classrooms before the start of classes, and written informed consent was obtained from all participants.

A structured data collection form, developed by the researchers based on a comprehensive literature review, was used to assess sociodemographic characteristics and lifestyle risk factors for NCDs. Content validity was ensured through expert review by researchers in nutrition and public health, who provided feedback on the relevance and clarity of the questions. The form was administered to students face-to-face, with each interview lasting approximately 15 min. The assessed lifestyle risk factors for NCDs included physical inactivity, low fruit and vegetable intake, alcohol consumption, tobacco use, being overweight or obese, and SSB consumption. Researchers also collected anthropometric measurements along with self-reported data on health, household characteristics, and living conditions.

2.1 Population

Students in the Nutrition and Dietetics program who were aged between 18 and 25 years, present during the classroom sessions and willing to complete the data collection form were included in the study. Students absent during data collection or failing to complete the form were excluded. In this study, purposive sampling (a type of non-probability sampling) was employed to target a specific population of interest—nutrition and dietetics students—due to their relevance to the study objectives. This approach was chosen to ensure that the sample represented students with a foundational understanding of health and nutrition, making them a critical population for assessing the prevalence and co-occurrence of non-communicable disease (NCD) risk factors. To ensure an adequate sample size, a power analysis was conducted. Based on an 80% power, 0.05 margin of error, medium effect size, and an anticipated 10% data loss, it was determined that at least 320 students were required to achieve statistical significance. Our goal was to include the entire population of nutrition and dietetics students while maintaining a robust sample size. Ultimately, a total of 485 students participated in the survey, with a response rate of 93%. The study included undergraduate students aged 18–25 years who provided informed consent. Measures were taken to reduce response bias, including anonymizing responses and emphasizing the voluntary nature of participation. While purposive sampling does not allow for generalization to all university students, it was appropriate for this study because the target population had specific characteristics critical to the research question. This method allowed us to focus on a group particularly relevant to the investigation of NCDs risk factors. Additionally, this sampling strategy was supplemented by a large sample size, which enhances the reliability of the findings within this population.

2.2 Questionnaire

Height (cm) and weight (kg) data were collected on the basis of self-reports, and BMI was calculated with the formula weight/height^2 to determine whether the participants were normal weight (≤ 24.9 kg/m^2), overweight (25.0–29.9 kg/m^2) or obese (≤ 30 kg/m^2) according to the World Health Organization (WHO) (11). Being overweight or obese was also considered a risk factor. The physical activity level (frequency of moderate-or high-intensity physical activity lasting at least 30 min without interruption) was measured daily, 5–6 days a week, 2–4 days a week and ≤ 1 day a week, and the activity levels were evaluated. A physical activity level of <150 min/week (12) was considered an insufficient level of physical activity and a risk factor. Smoking status was classified as active smoker, previous smoker but later quit, or nonsmoker. Active smoking is an accepted risk factor. Alcohol consumption was assessed by asking, "Have you drank alcohol at least once in the last month?" to assess current alcohol consumption. A positive answer to this question was considered to be the use of alcohol. The frequency of alcohol use was assessed by asking about the "frequency of consuming alcoholic beverages in the amount of one glass/glass of wine" and classifying it as almost daily, 4–6 days a week, 1–3 days a week, a few days a month, less, or not at all. No safe limit exists for alcohol consumption (13, 14); therefore, it was accepted as a risk factor.

The WHO recommends consuming more than 400 g of fruits and vegetables daily to improve overall health and reduce the risk of certain NCDs (15). In this study Food Frequency Questionnaire (FFQ) was used to assess fruit and vegetables intake. Low vegetable consumption was defined as consuming four or fewer portions per day (approximately 80 g per portion), while low fruit consumption was defined similarly. For sugar-sweetened beverage (SSB) consumption, individuals who consumed SSBs at least once per week were classified as being at risk, while those who never or consumed on occasion SSBs (a few days per month) were not. These thresholds were chosen to reflect dietary behaviors associated with increased NCD risk.

2.3 Statistics

Data were analyzed using SPSS version 26 (16). The prevalence and 95% confidence intervals (CIs) were calculated to identify the risk factors for NCDs according to sociodemographic characteristics. Differences between the measurement variables were examined using *t*-tests. Count-type variables were compared according to sociodemographic characteristics using chi-square tests.

A total of 128 potential combinations of the seven risk factors were initially analyzed (all possible combinations of the seven variables = $2^{\wedge 7}$). However, certain combination groups were excluded due to the absence of participants. Finally, 77 combinations were included in the analysis. Risk factors associated with the same period were considered as co-occurring. Having four or more risk factors was considered the threshold for identifying co-occurrence, and after this threshold, the rates suddenly decreased.

A multivariate logistic regression model was built to examine the odds ratios (ORs) and 95% CIs for the associations between the determined variables (sex, age, and education level) and four or more risk factors. For all analyses, statistical significance was set at $p < 0.05$.

3 Results

In total, 485 (86.6% women) students, comprising 265 (54.6%) from state universities and 220 (45.4%) from private universities, were included in this study, with a mean age of 21.21 ± 3.10 years. Overall, 23.5% were in their first year of university, 30.5% in their second year, 24.3% in their third year, and 21.8% in their fourth year. A total of 34.4% of the students lived in student houses during their university education, and 29.5% lived with their families.

Table 1 shows the prevalence and 95% CIs of the risk factors determined for the students. Overall, the most frequent risk factors were insufficient physical activity (89.2%), low fruit consumption (58.9%), low vegetable consumption (70.5%), alcohol consumption (44.5%), SSB consumption (41.1%), tobacco smoking (25.2%), and being overweight or obese (14.9%). Students living in their homes had a higher prevalence of tobacco (34.5%) and alcohol consumption (58.2%). Whereas, the prevalence of low vegetable (76.0%) and fruit (67.3%) content was higher among students living in dorms. Consumption of SSB was more prevalent among grade 1 students.

Table 2 shows the prevalence of co-occurrence for all combinations of the seven risk factors. Among the combinations, four risk factors (28.9%) had the highest prevalence, followed by three (22.6%) and two risk factors (18.3%). The highest prevalence (9.9%) observed for the combinations of risk factors was the co-occurrence of insufficient physical activity, low vegetable and fruit consumption, and SSB consumption (Figure 1). The observed prevalence in individuals with no risk factors was 0.9%, whereas that in individuals with all risk factors was 0.5%. When risk factors were examined alone, the highest prevalence was observed for insufficient physical activity (89.2, 95% CI: 86.1; 91.9), followed by low vegetable consumption (70.5, 95% CI: 66.3, 74.7), low fruit consumption (58.9, 95% CI: 54.2; 63.7), alcohol consumption (44.5, 95% CI: 40.6; 49.1), SSB consumption (41.1, 95% CI: 36.8; 45.5), tobacco smoking (25.2, 95% CI: 21.6; 29.3), and being overweight/obese (14.9, 95 %CI: 11.8; 18.3).

Overall, 51.2% (95% CI: 46.5; 56.0) of the students had four or more risk factors. Students with four or more risk factors were more likely to live in student houses (39.1%) and preferred fast food when eating out (59.5%). Moreover, among students with four or more risk factors, the prevalence of students with five or more hours of daily screen time was higher (60.1%) than that among those with fewer risk factors. The average daily water intake was higher (26.22 ± 11.14 mL) in students with three or more risk factors (Table 3).

The results of the multivariate logistic regression analysis for the co-occurrence of four or more risk factors for NCDs are shown in Table 4. The co-occurrence of four or more lifestyle risk factors was significantly higher among students at private universities (aOR: 2.01 95% CI: 1.2; 3.35), those living in student homes (aOR: 3.57 95% CI: 1.96; 6.5), and those with fast food preferences when eating outside (a0R: 2.53 95% CI: 1.62; 3.96). Conversely, a reverse association was observed between daily water intake (a0R: 0.98 95% CI: 0.96; 0.998) and four or more risk factors.

4 Discussion

This study evaluated the prevalence and co-occurrence of the modifiable lifestyle risk factors for NCDs among university students, considering their sociodemographic characteristics. A significant prevalence of co-occurring risk factors was observed, with over half of the participants presenting four or more risk factors. This underscores the compounded vulnerability of university students to lifestyle-related NCDs.

TABLE 1 Prevalence of risk factors for NCDs.

	Insufficient physical activity % (95% CI)	Tobacco smoking % (95% CI)	Alcohol consumption % (95% CI)	Low vegetable consumption % (95% CI)	Low fruit consumption % (95% CI)	Overweight or obesity % (95% CI)	SSBs consumption % (95% CI)
Total	89.2 (86.1; 91.9)	25.2 (21.6; 29.3)	44.5 (40.6; 49.1)	70.5 (66.3; 74.7)	58.9 (54.2; 63.7)	14.9 (11.8; 18.3)	41.1 (36.8; 45.5)
Gender							
Female	91.9 (88.8; 94.3)	24.8 (20.7; 29.2)	43.3 (38.5; 48.2)	70.0 (65.3; 74.4)	58.7 (53.8; 63.5)	12.2 (9.2; 15.7)	39.0 (34.1; 44.0)
Male	73.0 (60.3; 83.4)	28.1 (17.6; 40.8)	53.1 (40.2; 65.7)	74.2 (61.5; 84.5)	59.7 (46.4; 71.9)	32.8 (21.6; 45.7)	55.7 (42.4; 68.5)
Age							
18–20 years	90.2 (85.5; 93.7)	22.6 (17.3; 28.6)	43.8 (37.2; 50.5)	76.6 (70.4; 82.0)	61.3 (54.5; 67.7)	13.8 (9.6; 19.1)	50.0 (43.0; 57.0)
≥ 21 years	88.4 (83.8; 92.0)	27.4 (22.1; 33.3)	45.2 (39.0; 51.5)	65.2 (59.0; 71.1)	56.7 (50.4; 62.9)	15.9 (11.7; 20.9)	33.6 (27.7; 39.9)
Grade							
Grade 1	90.4 (83.4; 95.1)	21.1 (14.0; 29.7)	43.0 (33.7; 52.6)	69.6 (60.2; 78.0)	60.7 (51.0; 69.8)	14.2 (8.3; 22.0)	51.8 (42.1; 61.3)
Grade 2	89.7 (83.5; 94.1)	25.0 (18.3; 32.8)	40.5 (32.6; 48.9)	76.6 (68.8; 83.2)	57.6 (49.1; 65.8)	17.1 (11.4; 24.2)	41.7 (33.0; 50.8)
Grade 3	84.7 (77.0; 90.7)	24.6 (17.1; 33.4)	46.6 (37.4; 56.0)	76.9 (68.2; 84.2)	63.2 (53.8; 72.0)	13.6 (8.0; 21.1)	41.5 (32.5; 51.0)
Grade 4	92.4 (85.5; 96.7)	30.5 (21.9; 40.2)	49.5 (39.6; 59.5)	55.4 (45.2; 65.3)	53.5 (43.3; 63.5)	14.3 (8.2; 22.5)	28.0 (19.5; 37.9)
University							
Public	87.5 (83.0; 91.3)	22.3 (17.4; 27.8)	42.3 (36.2; 48.5)	69.9 (63.9; 75.5)	62.7 (56.5; 68.7)	16.7 (12.4; 21.7)	37.3 (31.4; 43.4)
Private	91.2 (86.7; 94.6)	28.6 (22.8; 35.1)	47.3 (40.5; 54.1)	71.2 (64.7; 77.1)	54.3 (47.5; 61.1)	12.8 (8.7; 18.0)	46.4 (39.2; 53.7)
Mother's education level							
Primary or secondary school	89.7 (84.3; 93.7)	22.3 (16.5; 29.0)	34.8 (27.9; 42.1)	70.2 (62.9; 76.8)	60.1 (52.5; 67.4)	14.7 (9.9; 20.6)	36.7 (29.6; 44.3)
High school or university	89.2 (85.0; 92.5)	27.2 (22.2; 32.6)	50.7 (44.8; 56.5)	71.1 (65.5; 76.2)	58.4 (52.5; 64.1)	15.3 (11.3; 19.9)	44.0 (38.1; 50.1)
Father's education level							
Primary or secondary school	90.8 (84.9; 95.0)	23.6 (16.9; 31.4)	38.9 (30.9; 47.4)	66.4 (58.0; 74.2)	58.6 (49.9; 66.8)	16.0 (10.4; 23.0)	37.5 (29.4; 46.2)
High school or university	88.1 (84.1; 91.4)	26.0 (21.3; 31.1)	46.8 (41.3; 52.4)	73.4 (68.2; 78.1)	59.3 (53.7; 64.7)	14.5 (10.9; 18.8)	43.0 (37.4; 48.8)
Residence during education							
Living with family	89.4 (83.1; 93.9)	18.2 (12.2; 25.5)	34.3 (26.5; 42.7)	64.5 (56.0; 72.4)	45.4 (37.0; 54.0)	14.8 (9.4; 21.7)	32.8 (24.9; 41.6)
Student house	87.9 (81.9; 92.4)	34.5 (27.3; 42.3)	58.2 (50.3; 65.8)	69.8 (62.1; 76.7)	61.5 (53.5; 69.0)	18.2 (12.6; 24.9)	39.9 (32.2; 48.0)
Dorm	90.3 (84.9; 94.2)	21.6 (15.8; 28.4)	40.3 (33.0; 48.0)	76.0 (68.9; 82.2)	67.3 (59.7; 74.2)	12.1 (7.6; 17.9)	48.5 (40.7; 56.3)

TABLE 2 Prevalence and co-occurrence patterns of risk factors for NCDs.

	Insufficient physical activity	Tobacco smoking	Alcohol consumption	Low vegetable consumption	Low fruit consumption	Overweight or obesity	SSB consumption	Observed prevalence (%)
7 (0.5%)	+	+	+	+	+	+	+	0.5
6 (5.6%)	+	+	+	+	+	+	–	0.2
	+	**+**	**+**	**+**	**+**	**–**	**+**	**3.9**
	+	+	+	–	+	+	+	0.2
	+	+	–	+	+	+	+	0.5
	+	–	+	+	+	+	+	0.9
5 (16.3%)	**+**	**+**	**+**	**+**	**+**	**–**	**–**	**5.1**
	+	–	–	+	+	+	+	0.5
	+	+	+	–	–	+	+	0.2
	–	+	+	+	+	+	–	0.2
	+	–	+	+	+	+	–	1.2
	+	–	+	–	+	+	+	0.2
	+	+	–	+	–	+	+	0.7
	+	+	+	–	+	–	+	0.2
	–	+	+	+	+	–	+	0.9
	+	–	+	+	+	–	+	4.8
	+	+	–	+	+	–	+	0.9

(Continued)

TABLE 2 (Continued)

	Insufficient physical activity	Tobacco smoking	Alcohol consumption	Low vegetable consumption	Low fruit consumption	Overweight or obesity	SSB consumption	Observed prevalence (%)
4 (28.9%)	+	+	+	+	–	–	–	1.8
	+	+	+	–	+	–	–	0.7
	+	+	+	–	–	–	+	0.9
	+	+	–	+	+	–	–	0.7
	+	+	–	+	–	+	–	0.5
	+	+	–	–	+	+	–	0.2
	+	+	–	–	+	–	+	0.5
	+	–	+	+	+	–	–	6.2
	+	–	+	+	–	+	–	0.9
	+	–	+	+	–	–	+	2.5
	+	–	+	–	+	+	–	0.5
	+	–	+	–	+	–	+	0.7
	+	–	+	–	–	+	+	0.5
	+	–	–	+	+	+	–	0.9
	+	**–**	**–**	**+**	**+**	**–**	**+**	**9.9**
	+	–	–	+	–	+	+	0.2
	+	–	–	–	+	+	+	0.2
	–	+	+	+	–	+	–	0.2
	–	+	+	–	+	+	–	0.2
	–	+	+	–	–	+	+	0.5
	–	+	–	+	+	+	–	0.2
	–	–	–	+	+	+	+	0.2
	–	–	+	+	+	–	+	0.2

(Continued)

TABLE 2 (Continued)

	Insufficient physical activity	Tobacco smoking	Alcohol consumption	Low vegetable consumption	Low fruit consumption	Overweight or obesity	SSB consumption	Observed prevalence (%)
3 (22.6%)	–	–	–	+	+	–	+	0.2
	–	–	+	–	+	+	–	0.2
	–	–	+	+	–	–	+	0.2
	–	+	+	–	–	–	+	0.2
	–	+	+	–	–	+	–	0.2
	–	+	+	+	–	–	–	0.2
	+	–	–	–	–	+	+	0.7
	+	–	–	–	+	–	+	0.7
	+	–	–	+	–	–	+	2.3
	+	–	–	+	–	+	–	0.9
	+	**–**	**–**	**+**	**+**	**–**	**–**	**9.7**
	+	–	+	–	–	–	+	0.9
	+	–	+	–	+	–	–	1.6
	+	–	+	+	–	–	–	2.8
	+	+	–	–	–	–	+	0.2
	+	+	+	–	–	–	–	0.7
	+	+	–	–	+	–	–	0.7
	+	+	–	+	–	–	–	0.2

(Continued)

TABLE 2 (Continued)

	Insufficient physical activity	Tobacco smoking	Alcohol consumption	Low vegetable consumption	Low fruit consumption	Overweight or obesity	SSB consumption	Observed prevalence (%)
2 (18.3%)	–	–	–	–	–	+	+	0.5
	+	+	–	–	–	–	–	0.7
	–	+	+	–	–	–	–	0.2
	–	–	+	+	–	–	–	0.5
	–	–	–	+	+	–	–	1.2
	–	–	–	–	+	+	–	0.2
	+	–	–	–	–	–	+	2.5
	–	+	–	–	–	–	+	0.2
	+	–	+	–	–	–	–	1.4
	–	+	–	+	–	–	–	0.2
	+	**–**	**–**	**+**	**–**	**–**	**–**	**5.5**
	+	–	–	–	+	–	–	3.5
	+	–	–	–	–	+	–	1.4
	–	–	+	–	–	+	–	0.2
	–	+	–	–	+	–	–	0.2
1 (7.0%)	–	–	–	+	–	–	–	0.9
	–	–	+	–	–	–	–	0.2
	+	**–**	**–**	**–**	**–**	**–**	**–**	**6.2**
0 (0.9%)	–	–	–	–	–	–	–	0.9

The symbol '+' represents the presence of the factor, and '–' represents the absence of the factor. The bold values represent $p < 0.005$ and indicate statistical significance.

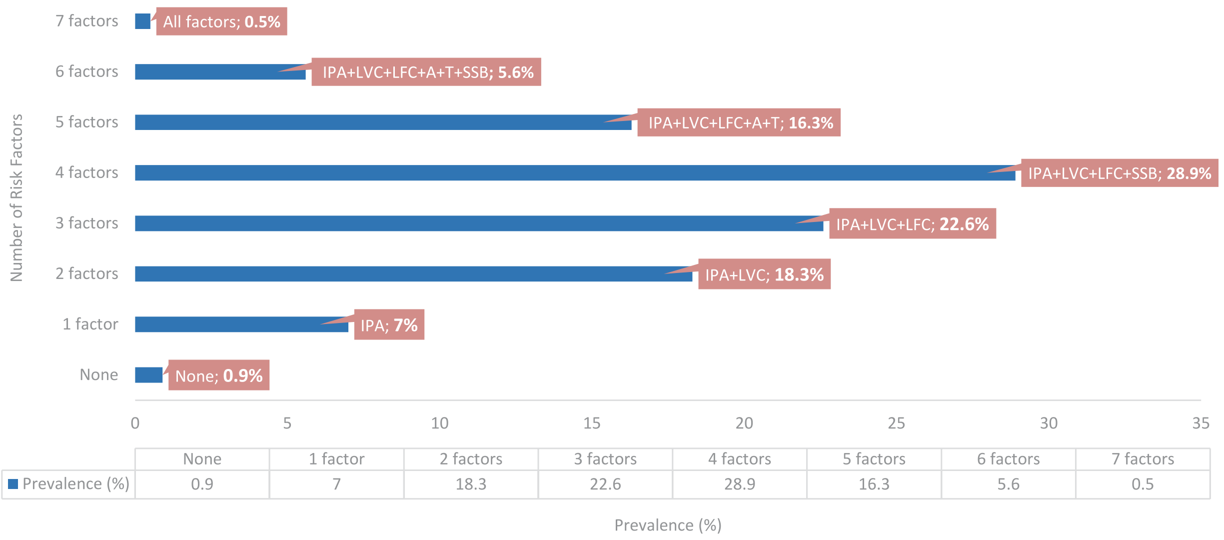

FIGURE 1
Prevalence of risk factors co-occurrence and most common combinations. IPA, insufficient physical activity; LVC, low vegetable consumption; LVF, low fruit consumption; A, alcohol consumption; T, tobacco smoking; SSB, sugar-swetened beverages consumption.

4.1 Insufficient physical activity

The prevalence of insufficient physical activity was alarmingly high at 89.2%, consistent with global trends among university students. This rate is concerning, as sedentary behavior is a well-established risk factor for non-communicable diseases (NCDs), including cardiovascular disease, diabetes, and certain cancers (17). Approximately 81% of adolescents worldwide fail to meet physical activity guidelines and more girls inactive than boys in most countries, reflecting a pervasive issue (18). In 2012, a related study investigating the impact of physical inactivity on major NCDs globally revealed striking finding: The research projected that eliminating physical inactivity could result in a median increase in life expectancy of 0.68 years worldwide, 0.63 years in Europe, and 1.06 years in Turkey (17). Academic life, characterized by prolonged sedentary behavior, screen time, and limited access to recreational facilities, likely contributes to these patterns particularly for university students (19). These findings emphasizes the critical need for specific solutions to promote physical activity among university students. Policymakers and university administrators must prioritize accessible fitness facilities, structured physical activity programs, and campus-wide campaigns to encourage active lifestyles.

4.2 Low fruit and vegetable consumption

In this study, roughly 7 in 10 students had low vegetable consumption, and 6 in 10 students had low fruit consumption. A study assessing university students from 26 countries across America, Asia, and Africa and reported a great prevalence (82.8%) of inadequate fruit and vegetable consumption, underscoring the need for improved intake among university students globally which aligns with these findings (20). University students were more likely to have a low frequency of fruit and vegetable consumption suggesting that research conducted among young adults, particularly university students, has reported a high prevalence of insufficient fruit and vegetable consumption. For instance, rates were 85.2% in Brazil (21), 95% in Germany (22), 73.6% in Saudi Arabia (23), and 70% in the UK (24). Financial constraints, time limitations, and easy availability of processed foods often dictate dietary habits of university students (5). Adequate fruit and vegetable intake is crucial for providing necessary nutrients and dietary fiber, vitamin and minerals which protect against NCDs (25). Policies to subsidize healthy food options, increase access to fresh produce on campuses, and integrate nutrition education into health programs are essential.

4.3 SSB consumption

The excessive intake of SSBs by university students is a major public health concern. Since added sugars in beverages raise blood glucose and insulin levels, which may lead to an increased risk of type 2 diabetes (26). In this study, over 2 in 5 students reported frequent SSB intake. Younger populations are particularly susceptible due to the affordability, availability, and aggressive marketing of sugary drinks (27). In the United States, the National Longitudinal Study of Adolescents and Adults Health reported a high prevalence of SSB consumption, with 87.3% of participants having consumed SSBs in the previous week and 47.8% consuming eight or more such beverages. Additionally, young adults who consume sugar-or artificially sweetened beverages tend to have a higher cumulative disease burden.

From another perspective, there has been growing interest in encouraging water consumption as a strategy to reduce SSB intake, with the expectation that increasing water consumption can decrease SSB consumption by replacing it. Notably, students with four or more risk factors were found to have lower water intake, highlighting an inverse association. This suggests that encouraging higher water consumption could be an effective approach to reducing SSB intake. Supporting this, a systematic review concluded that promoting water intake can significantly

TABLE 3 Comparison of sociodemographic variables by the presence of four or more risk factors for NCDs.

		Three or less risk factors	Four or more risk factors	p
Overall		48.8% (44.0; 53.5)	51.2% (46.5; 56.0)	
Age ($n = 443$)	18–20 years	90 (41.7%)	114 (50.2%)	0.071
	≥21 years	126 (58.3%)	113 (49.8%)	
Gender ($n = 443$)	Female	192 (88.9%)	193 (85.0%)	0.228
	Male	24 (11.1%)	34 (15.0%)	
University ($n = 443$)	Public	131 (60.6%)	122 (53.7%)	0.142
	Private	85 (39.4%)	105 (46.3%)	
Grade ($n = 443$)	Grade 1	50 (23.1%)	59 (26.0%)	0.424
	Grade 2	57 (26.4%)	64 (28.2%)	
	Grade 3	55 (25.5%)	62 (27.3%)	
	Grade 4	54 (25.0%)	42 (18.5%)	
Residence ($n = 432$)	Living with family	74 (34.9%)	52 (23.6%)	**0.026**
	Student house	64 (30.2%)	86 (39.1%)	
	Dorm	74 (34.9%)	82 (37.3%)	
Frequency of eating outside ($n = 433$)	Less than once a week	83 (39.2%)	45 (20.4%)	**<0.001**
	1–3 times per week	87 (41%)	102 (46.2%)	
	Four or more times a week	42 (19.8%)	74 (33.5%)	
Preference of eating outside ($n = 431$)	In dining hall or one-pot meals	103 (61.7%)	64 (38.3%)	**<0.001**
	Fast food	107 (40.5%)	157 (59.5%)	
Daily screen time ($n = 434$)	Less than 5 h	107 (50.7%)	89 (39.9%)	**0.020**
	5–9 h	97 (46.0%)	116 (52.0%)	
	10 h or more	7 (3.3%)	18 (8.0%)	
		Mean ± SD	Mean ± SD	
		4.77 ± 2.17	5.36 ± 2.38	**0.007**
Daily water intake (ml/kg) ($n = 420$)		26.22 ± 11.14	23.96 ± 11.08	**0.038**

SD, Standard deviation. h, hours. The bold values represent $p < 0.005$ and indicate statistical significance.

decrease SSB consumption (28). These findings suggest that public health messages for young adults should include warnings about both sugar-sweetened and artificially sweetened beverages (29). To mitigate these risks, public health measures such as taxation on sugary drinks, educational campaigns, increasing the availability of healthier beverage options like water should be prioritized.

4.4 Alcohol consumption and tobacco use

Tobacco smoking and alcohol consumption are prevalent risk behaviors among university students, with this study reporting rates of 25.2% for smoking and 44.5% for alcohol use. Similarly, a study conducted among university students in China reported that 29.8% of students smoked or used e-cigarettes (30). These behaviors are often influenced by peer pressure, stress, and normalization within student cultures. University students aged between 18 and 25 are at greater risk of initiating tobacco smoking due to the transition point from high school to college (31) Universities, therefore, ought to allocate resources toward the prevention and treatment of students at risk for alcohol, smoking and drug use disorders to reduce the effects on their academic performance and mental health throughout their university experience (31).

4.5 Co-occurrence of risk factors

A significant prevalence of risk factor co-occurrence was found in more than half of the research population (51.2%) which had four or more risk factors. Moreover, a reverse association was observed between daily water intake and four or more risk factors. Previous studies have shown that risky behaviors commonly co-occur, with 52% in the USA (32), 55% in the Netherlands (33), 59% in Brazil (17), and 68% in England (34). Accordingly, four lifestyle risk variables (inadequate fruit and vegetable intake, smoking, alcohol consumption, and poor physical activity) were studied, focusing on the co-occurrence and clustering profiles of cardiovascular lifestyle risk factors among adults in West Africa. The prevalence of two or more cardiovascular lifestyle risk factors co-occurring was 46.4% (35).

This study revealed that the most common combination of risk factors was the co-occurrence of insufficient physical activity, low vegetable and fruit consumption, and SSB consumption, indicating a

TABLE 4 Multivariate logistic regression analysis of sociodemographic characteristics associated with the co-occurrence of four or more risk factors ($n = 407$; $R^2 = 0.164$).

	aOR (95% CI)	*p*
University$^{\text{Public}}$		
Private	2.01 (1.2; 3.35)	**0.008**
Grade$^{\text{Grade 1}}$		0.505
Grade 2	0.98 (0.53; 1.79)	0.941
Grade 3	1.00 (0.53; 1.89)	1.00
Grade 4	0.62 (0.28; 1.35)	0.226
Age$^{\text{18–20 years}}$		
>21 years	0.98 (0.59; 1.65)	0.946
Gender$^{\text{Female}}$		
Male	1.67 (0.88; 3.19)	0.117
Mother's education level$^{\text{high school or university}}$		
Primary or secondary school	0.73 (0.47; 1.14)	0.169
Residence$^{\text{Living with family}}$		**<0.001**
Student house	3.57 (1.96; 6.5)	**<0.001**
Dorm	2.47 (1.34; 4.56)	**0.004**
Daily screen time$^{\text{Less than 5 h}}$		0.459
5–9 h	1.25 (0.80; 1.95)	0.329
10 h or more	1.67 (0.61; 4.62)	0.322
Daily water intake (ml/kg)	0.98 (0.96; 0.998)	**0.031**
Preferences of eating outside $^{\text{In dining hall or one-pot meals}}$		
Fast food	2.53 (1.62; 3.96)	**<0.001**

OR, odds ratio; 95% CI, confidence interval 95%. References are shown as superscripts. The bold values represent $p < 0.005$ and indicate statistical significance.

group with an increased risk of NCDs. Similarly, a systematic review revealed an especially high prevalence of insufficient physical activity and low fruit and vegetable intake (36). These findings are consistent with data from other countries, including the United States (37), where low fruit and vegetable intake and physical inactivity were the most common co-occurring behaviors. Most studies have focused on adult populations at the center, with few studies considering younger adults or students. Notably, the primary limitation of the studies with co-occurrence was the fluctuating cutoff values for risk attitudes. These limitations make it harder to compare studies and are likely to contribute to the observed variability in most data.

In this study, students with four or more risk factors are less likely to live with their families compared to those with fewer risk factors. This finding aligns with literature suggesting that students living away from family often experience fewer home-cooked meals due to factors like limited cooking facilities and time constraints, thus increasing their susceptibility to poor dietary patterns (38).

This study revealed an inverse relationship between daily water intake and the presence of four or more risk factors. Although no clustered studies exist on the co-occurrence of risk factors for NCDs and daily water consumption, one prospective study reported that higher water intake was associated with a lower risk of mortality (39). Similarly, a systematic review of prospective cohort studies demonstrated that higher total water consumption was associated with a decreased risk of cardiovascular diseases mortality (40). Because proper hydration and water consumption are required for important physiological and metabolic functions, understanding the relationship between water consumption and NCD risk factors is critical for policymakers.

Research on this topic is exceptionally heterogeneous, with different approaches for defining and assessing risk factors, and no consensus on which risk factors typically occur together (41). Therefore, studies that cluster and comprehend the different risk factors for NCDs and include different age groups, especially among university students, are needed.

4.6 Strengths and limitations

This study's strengths include its comprehensive analysis of multiple NCD risk factors and their co-occurrence, addressing a critical gap in the literature where studies on the co-occurrence of risk factors among university students are notably scarce. By examining variables like residence, eating habits, and screen time, it provides valuable insights into clustering behaviors. The use of robust statistical methods enhances reliability, while the focus on an underrepresented population contributes to global NCD prevention efforts.

This study had several limitations. First, its cross-sectional design limits the ability to establish causal relationships. The dependence on self-reported data may lead to recall and social desirability biases, as participants could understate undesirable actions or overstate behaviors deemed socially acceptable. To mitigate this, anonymity was ensured during data collection. Nonetheless, the absence of a fully standardized and formally validated questionnaire may affect the accuracy and reliability of self-reported behaviors. Furthermore, the purposive sampling method limits the generalizability of the findings to university students from institutions with different cultural, geographical, and academic characteristics. The predominance of female participants and the inclusion of only two universities, one private and one public, may also have introduced selection bias. Future research using randomized sampling across diverse settings and validated instruments is needed to confirm and expand upon these findings.

5 Conclusion

Our findings highlight the urgent need for university-based interventions targeting physical activity and dietary habits to reduce the risk of non-communicable diseases (NCDs) among students. Programs that promote structured physical activity, collaborative efforts between university administrations and public health bodies to implement wellness programs, provide healthier, nutritious, affordable dining options on campus and educate students on healthy lifestyle choices could play a pivotal role in mitigating these risks. The identification of multiple risk factors in many students underscores the necessity of early action. Intervening during university years not only fosters healthier habits that persist into adulthood but also contributes to healthy aging, reducing the burden of NCDs later in life. Additionally, such proactive measures can lower both the direct costs (e.g., treatment expenses) and indirect costs (e.g., lost productivity) for healthcare systems. Future research should explore diverse clusters of risk factors and comprehensive data to refine these strategies and optimize their impact on university populations.

Data availability statement

The data that support the findings of this study are available from the authors, upon reasonable request.

Ethics statement

The studies involving humans were approved by Aydin Adnan Menderes University Faculty of Medicine Clinical Research Ethics Committee. The studies were conducted in accordance with the local legislation and institutional requirements. The participants provided their written informed consent to participate in this study.

Author contributions

IC: Conceptualization, Investigation, Software, Writing – original draft. SG: Conceptualization, Investigation, Methodology, Supervision, Writing – review & editing.

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Conflict of interest

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

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2024.1484164/full#supplementary-material

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EDITED BY
Mo Salman,
Colorado State University, United States

REVIEWED BY
Anubhuti Dubey,
Deen Dayal Upadhyay Gorakhpur University,
India
Ezgi Fındık,
Ankara University, Türkiye

*CORRESPONDENCE
Anmut Endalkachew Bezie
✉ anmut1216@gmail.com

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Work-related burnout and its associated factors among kindergarten teachers: a multi-center cross-sectional study in Ethiopia

Anmut Endalkachew Bezie[1]*, Girum Tareke Zewude[2], Amensisa Hailu Tesfaye[3,4], Asmare Asrat Yirdaw[5], Alebachew Bitew Abie[6] and Giziew Abere[3]

[1]Department of Occupational Health and Safety, College of Medicine and Health Sciences, Wollo University, Dessie, Ethiopia, [2]Department of Psychology, Wollo University, Dessie, Ethiopia, [3]Department of Environmental and Occupational Health and Safety, Institute of Public Health, College of Medicine and Health Sciences, University of Gondar, Gondar, Ethiopia, [4]Institute for Sustainable Futures, University of Technology Sydney, Sydney, NSW, Australia, [5]Department of Environmental Health, College of Medicine and Health Science, Arba Minch University, Arba Minch, Ethiopia, [6]Department of Environmental Health, College of Medicine and Health Sciences, Wollo University, Dessie, Ethiopia

Introduction: Work-related burnout is a state of severe physical and mental fatigue and exhaustion resulting from unmanaged prolonged work stress. Kindergarten teachers are at higher risk of work-related burnout compared to other teaching professionals, due to their dual roles as both caregivers and educators for young children. The demands and stresses of these combined responsibilities, coupled with factors such as low salaries, contribute to an increased risk of burnout in this population. Therefore, this study aimed to investigate work-related burnout prevalence and associated factors among kindergarten teachers in Dessie City, Northeast Ethiopia.

Methods: An institution-based, cross-sectional study was conducted in March and April of 2024. A sample of 422 kindergarten teachers was recruited using simple random sampling techniques. A standardized, self-administered Copenhagen Burnout Inventory was utilized to measure work-related burnout. We used EpiData V4.6 and SPSS V26 for data entry and analysis, respectively. A bivariable logistic regression analysis (p-value < 0.2) was performed to find factors associated with work-related burnout. Variables found significant in the bivariable analysis were then exported into a multivariable logistic regression model to identify statistically significant variables at a p value < 0.05 and a 95% confidence interval.

Results: The response rate was 95% (401/422). 97.5% of the participants were female, and the participants' mean (standard deviation) age was 28.5 years (SD ± 5.8). In the past 6 months, the total prevalence of WRB was 39.7% [95% CI (34.8, 44.6)]. Work–family conflicts [AOR: 2.81; 95% CI (1.77, 4.45)], working conditions [AOR: 1.63; 95% CI (1.03, 2.56)], perceived stress [AOR: 1.91; 95% CI (1.21, 3.01)], job dissatisfaction [AOR: 1.75, 95% CI (1.10, 2.79)], and kindergarten type [AOR: 1.66; 95% CI (1.02, 2.68)] were factors significantly associated with WRB.

Conclusion: According to this study, a significant number of kindergarten teachers were affected by burnout due to their working nature. To address this issue, interventions focused at reducing work–family conflicts, increasing job satisfaction, and improving the school environment are necessary to support teachers' well-being, benefiting both young students and the broader educational landscape.

KEYWORDS

work-related burnout, kindergarten teachers, burnout, children, Ethiopia

Introduction

Work-related burnout is a state of severe physical and mental fatigue and exhaustion resulting from unmanaged prolonged work stress (1). It is the result of extensive and prolonged work-related stress, characterized by emotional exhaustion leading to chronic fatigue and a lack of emotional energy; cynicism, which refers to mentally detaching oneself from work and clients; and a reduction in professional performance such as reduced teaching ability, incompetence, and ineffectiveness (2). Work-related burnout (WRB) is a significant public health challenge in the education sector globally. It develops gradually over time, often without the affected individual being fully aware of it (3). Burnout has implications beyond health; it affects a teacher's passion and commitment and ultimately compromises their ability to meet the needs of their students (4).

Kindergarten (KG) education is an educational process that spans the years from birth to elementary school. It plays a significant role in children's academic formation, social and emotional development, and health promotion, which requires the physical and mental effort of the teachers in order to provide care and education for children up to the age of primary education (5). It is a specific focus area of Sustainable Development Goals (SDGs), particularly stated under SDG 4, which outlines its objective to "ensure that by 2030, all children have access to quality early childhood care, and pre-primary education"; by doing this, they will be ready for primary school and serve as a foundation for learning and development throughout their lives (6). It is important in terms of providing stimulating environmental opportunities that are appropriate for children's development and individual characteristics, as well as supporting children's physical, mental, emotional, and social development (7).

During their preschool period, kids begin to explore, learn, and interact with their surroundings and develop habits and behaviors that fit the social structure of the culture they live in. At this age, KG teachers are essential in giving the intentional guidance that a kid needs at home, at school, and in their social life (8). Mini (9) explained that since today's children are the citizens of tomorrow's world, it is necessary to empower them with knowledge and resources to meet their basic needs to enable them to grow to their full potential, as they are the foundation for national development. Therefore, to meet this goal, maintaining the psychological wellbeing of KG teachers is crucial to treating students, making them productive, and being responsible members of the future generation. However, despite Ethiopia is currently giving attention to early childhood development and has created the National Policy Framework, Strategic Operational Plan, and Guidelines for the implementation of high-quality programs for early childhood care and education, the mental health of KG instructors is not well-maintained (10). Presently, the Ministry is also promoting the involvement of private organizations and individuals to fund KG education through missions, non-profits, private individuals, religious institutions, and other organizations that are reopening in urban areas. However, teaching young children in Ethiopia is still regarded as one of the lowest professions due to low pay and unfavorable working conditions that drain their energy and lead to fatigue, which results in teachers losing interest in teaching as a career path and running the risk of burnout. KG teachers work is one of the most demanding and stressful jobs due to their daily responsibilities and frustrations, which often result in physical and mental demands that cause tiredness (5). Their work includes daily hassles like bringing homework, showing up early to school, and staying up late at home, all of which are part of the extremely tough and demanding job that leads to burnout (11).

Apart from this, they have a lot of noticeable and invisible responsibilities that must be fulfilled within a set amount of time that cause them less flexibility and tight work, which is a pertinent cause of burnout (12). The profession by itself needs a responsive caregiver, a competent listener, observer, actor, and topic expert since children who are not physically or mentally mature require the patience and enthusiastic nature of the teachers. However, fulfilling these tasks and managing the emotions accompanying them can be very stressful for early educators, especially if they need the proper training and support (13).

Instructors who experience burnout may be too emotionally exhausted to form close relationships with their students, making it difficult for them to serve as positive role models and communicate with them effectively (14). Children may be bored with learning or not motivated to participate in classroom activities where teachers are exhausted and lack the energy or enthusiasm to encourage their learning, and children may feel the teacher lacks confidence in their teaching (15).

KG teachers' burnout is thought to have a significant effect on how effectively and successfully they carry out their duties and is reflected on their students, students' parents, families, and administrators as well (16). This suggests that taking care of teachers' well-being is not merely an individual concern but has broader implications for the overall educational landscape. Compared to higher-ranking elementary-grade teachers, kindergarten instructors have a yearly turnover rate of 50% or move to another school as a result of working conditions, including burnout at work (17, 18).

From a public health point of view, burnout impacts educators, young children, and the community at large. It caused them lower self-esteem, diminished focus, psychological symptoms like lowered self-control, anger, alienation, anxiety, and depression, and physical symptoms like headaches, stomach issues, elevated blood pressure, sleeplessness, musculoskeletal pain, and increased vulnerability to infections (19, 20). KG teachers WRB is a predictor of performance declines, absenteeism (21, 22), voice disorder (23), hurt teacher-child interaction (24), and reduced students' autonomous motivation (25). A teacher's burnout can have a detrimental impact on a number of areas of the classroom environment, including constructive relationships with students or the giving of constructive feedback, pessimism, carelessness, demonization, change resistance, a lack of creativity in instruction, and an unjustified absence (26). The peculiar nature of teaching among KG teachers makes them highly exposed to burnout than other working populations. Prevalence of burnout among KG teachers in China (53.2%) (27), Germany (24.4%) (28), and Greece, KG teachers experience slightly more feelings of emotional exhaustion and depersonalization (29).

Previous research has reported key determinants of WRB such as low job satisfaction (30), KG type (31), working hours per day (32), educational background (33), organizational climate (34), poor or adverse working conditions (35), work stress (36, 37), lack of social support (38, 39), interpersonal conflict (40), and low salaries (27). In addition, previous research has identified various sociodemographic factors associated with burnout among teachers (41–43). For example, female educators are more stressed and burned out than male educators (41, 44). Teachers who are less experienced in their job have a decreased positive work mindset that leads to a serious intention to leave their current employment due to burnout at work (45). Moreover, previous existing literature also shows job stress is positively related to burnout, which means that the higher the stress, the higher the burnout (46). The more teachers gain social support, the less burnout they experience (47) and job satisfaction is protective for burnout (30, 48). Work relationships, role conflict and ambiguity, and a lack of developmental opportunities lead to increased burnout (49). High job demands (50), poor leadership (51), work–family conflicts (52–55), conflict with colleagues, and low decision-making autonomy were factors for burnout (41). However, to date, the issue of WRB has received little attention from the management of the education and teaching sectors.

Work-related burnout is currently the leading occupational health problem affecting service delivery in the education system, and it has become the focus of numerous studies around the world (56). However, it appears that the management of the education and teaching sectors has given little attention to the issue of psychological hazard. There is a paucity of study on Burnout among KG teachers in the study setting and in Ethiopia at large. Moreover, identifying KG teachers' WRB level helps in increasing their mental and functional performance to satisfy students' needs during this vital educational stage. Therefore, the purpose of this study was to find WRB prevalence and associated variables among KG teachers in Dessie, northeastern Ethiopia. This study will provide important evidence about the level of burnout and associated variables among KG teachers, enabling the implementation of interventions or policy changes that will enhance teachers' professional well-being and ultimately improve the quality of education. Moreover, the study's findings could be pivotal in shaping educational policies and support programs specifically tailored to Ethiopian kindergarten teachers. Given the unique socio-cultural and economic factors in Ethiopia, it is crucial to address the specific challenges faced by educators, particularly in early childhood education.

Theoretical framework of current study

The theoretical framework of the current study was developed with the help of a literature review. It helped in the extraction of pre-school-level variables and exploring their link with work-related burnout among kindergarten teachers. Different theoretical frameworks that are essentially important for the development and occurrence of burnout were retained to represent the backup theory of burnout (57). Structural theory in occupational health focuses on how an organization's structure and culture impact employee well-being and burnout. Burnout occurs when rigid organizational structures, unclear roles, low job recognition, inadequate support, and high-pressure cultures and infective coping strategies create excessive stress and emotional exhaustion. This leads to decreased job performance and well-being, necessitating structural changes to support employees effectively (58). Demand-resource theory states that burnout occurs when the demands of work exceed the available resources. This imbalance initially causes fatigue, and if it continues over time, it leads to chronic fatigue and ultimately burnout. For kindergarten teachers, such demands may include work stress, working conditions, managing large class sizes, addressing the emotional needs of young children, and balancing the heavy workload of teaching and administrative responsibilities (59). In contrast, job resources, including job satisfaction, social support, supportive management, access to teaching materials, and a positive work environment, mitigate the impact of these demands, lowering the risk of burnout (60). Social Exchange Theory posits that burnout arises in the social and interpersonal context of the work organization, which is when there is unreciprocated effort at the organizational level or when employees' expectations regarding the nature of their exchange with the organization are not met (61). Under this theory, perceived value in the exchange can enhance job satisfaction; lack of reciprocity in support may lead to work–family conflicts; work misconduct issues; unmet expectations can cause to intentions to leave; and poor exchanges can contribute to dissatisfaction and burnout. Social cognitive theory: self-efficacy, self-confidence, and self-concept are the three main factors that leads to the evolution and development of burnout, and it arises when an employee has questions about their own or their group's effectiveness to accomplish work-related goals (58). Under this theory, taking early childhood care training can enhance self-efficacy, impacting how teachers manage stress while their job satisfaction may be affected by their perceptions of their effectiveness and self-concept. By integrating these theoretical perspectives, this study aimed to explore how organizational, work-related and personal factors contribute to burnout among kindergarten teachers. The development of our instrument is grounded in these frameworks, ensuring a comprehensive assessment of both work-related factors [work family conflicts, working condition, job demands and recognition, role clarity, job satisfaction, perceived stress, social support, taking early childhood care training, and individual-level (sociodemographic factors) contributors to burnout].

Materials and methods

Study design, period, and setting

An institution-based, cross-sectional study was conducted from March 4 to April 6, 2024, in Dessie, which is a multiethnic city in northeastern Ethiopia. It is located 488 kilometers from Bahirdar, the regional city of Amhara, and 401 kilometers from the capital city of Ethiopia, Addis Ababa. Dessie City has a total of 78 kindergarten schools: 49 private and 29 public. The number of educators in private kindergartens is above 395 and 240 in public kindergartens.

Population and eligibility criteria

The source population includes all kindergarten teachers in Dessie City and the study population was Teachers who had a minimum of 6 months of experience in the selected KG schools. However, participants who were on sick leave, maternity leave, or annual leave during the time of data collection were excluded in the study.

Sample size determination and sampling procedure

The sample size was calculated using a single population proportion formula with the following assumptions: Proportion ($p = 0.5$), because no previous study has been conducted among kindergarten teachers in Ethiopia. Margin of error ($d = 5\%$) and Z-score ($Z\alpha/_2 = 1.96$) corresponding to 95% of the confidence interval. $n = \left(Z\alpha/_2\right)^2 \frac{\left[p\left(1-p\right)\right]}{d^{2.}}$

$$n = \left(1.96\right)^2 \left[0.5\left(1-0.5\right)\right] / \left(0.05^2\right) = 384$$

By considering a 10% non-response rate, the total sample size (n) was: $n = 384^* \ 0.1 + 384 = 38.4 + 384 = 422$.

A sample was taken from Dessie City, with a total of 78 kindergarten schools: 49 private and 29 public. For private schools, the number of teachers is 6–24, and for public schools, it is 3–44 per school. We selected 35 private and 25 governmental kindergarten school teachers using simple random sampling. Thirty five selected private kindergarten schools have a total of 315 and 25 selected public schools have a total of 219; the overall number of teachers was 534. A simple random sampling procedure (lottery method) was used to select preschool teachers. To allocate a representative sample in each KG school, a proportional allocation to the size of the school teacher was used.

Measurement of variables and definition of terms

Work-related burnout: was measured using a Copenhagen Burnout Inventory with seven items. The presence or absence of work-related burnout was concluded with the average total score of 50, from which $< 50 = 0$ (no burnout) and $\geq 50 = 1$ (burnout) (1, 62).

Kindergarten teachers: are professionals who focus on providing care and education to children up to the age of primary education in public, non-public, or for-profit centers or schools that play a significant role in social development and health promotion (5).

Alcohol drinking: a kindergarten teacher who drinks any alcoholic drink at least two times per week (63).

Khat chewing: a kindergarten teacher who chewed khat three times a week for the last 6 months prior to the study (64).

Cigarette smoking: individuals who smoked at least one stick of cigarettes per day for the 6 months prior to the study (65).

Physical exercise: a kindergarten teacher who performed physical exercise at least two times per week for 30 min (66).

Job demand and role conflict with four items; job recognition and role clarity with three items on a 5-point Likert scale and dichotomized by their mean (67).

Job satisfaction: 10 items from a general job satisfaction scale score of 10–31 were satisfied, and 32 or above were dissatisfied (68).

Social support was measured by a 3-item Oslo Social Support Scale in which poor (3–8), moderate (9–11), and strong social support (12–14) (69).

Work–family conflicts were measured with 10 items on a seven-point Likert scale and dichotomized by mean (70).

Working conditions were measured by five items related to the conduciveness of class room conditions and school facilities (staff rooms, recreation centers, toilets, internet access, etc.), collegial relationships among teachers, and the number of kids in a class room, and dichotomized by their mean value (71).

Perceived work stress was measured by 14 items with five Likert scales and dichotomized as greater than or equal to 28 high and below 28 as low (72).

Turnover intention was measured by six items on a five-point Likert scale and dichotomized as below 18, which indicates an intention to stay, while a total score above 18 indicates an intention to leave (73).

Data collection tools and procedures

An anonymous, standard, self-administered questionnaire was used to collect the data. The development process of these instruments began with the selection of constructs, with a thorough literature review as measuring constructs relevant to this study. After that, each instrument's items were carefully selected based on their relevance to the specific objective of this study. The selection criteria are aligned with the theoretical frameworks. Once suitable instruments are identified, we have adapted to fit the cultural, social, and linguistic context of the study to our study population. Then the number of items from each instrument was chosen to measure specific constructs and communicating with health professionals, school teachers, and kindergarten teachers during the pretest to ensure the relevance of the questioner to measure the intended outcomes. Moreover, to ensure the tools' reliability, we test internal consistency of each construct.

The Copenhagen burnout inventory

The CBI was employed to evaluate WRB, which has been used and verified in previous research (74, 75). It is a universal tool that has been translated into multiple languages and is presently being utilized in numerous countries to assess risk factors related to the workplace (1). The CBI classifies burnout into three subscales, such as burnout related to personal, work, and client (76, 77). However, in the current study, only work-related burnout was investigated. WRB was made up of seven items. The validity and dependability of CBI in Africa were examined among Nigerian resident physicians, and the results revealed the highest internal consistency (78). This instrument is locally tailored to Ethiopia and has demonstrated excellent dependability in earlier research (79, 80). The instrument's response format and scoring system are as follows: each of the seven elicited items has a five-point Likert scale, with very low/never = 1, low/rarely = 2, somewhat/sometimes = 3, high/often = 4, and very high/always = 5. Following the data collection, for negative items, each Likert scale label was reverse-coded using one of the following formats: 1 = 5, 2 = 4, and 3 = 3. Then each Likert scale label was recoded using the following formats: 1 = 0, 2 = 25, 3 = 50, 4 = 75, and 5 = 100, and then all the items were summed. Then, each choice was given one of the following weighted percentages: never (0%), seldom (25%), sometimes (50%), often (75%), and always (hundred percent) (77, 81). In this study the internal dependability (Chronbach's alpha) was 0.82.

The Copenhagen psychosocial questionnaire

It is a standardized universal instrument with a five-point Likert scale intended to evaluate workplace health promotion and psychosocial circumstances (67). It is free and available to the public and can be applied to any kind of workplace. In Ethiopia, these tools are customized and extensively used in different working groups with good dependability (79, 80). We use the job demands scale from the Copenhagen Psychosocial Questionnaire to quantitatively assess specific aspects of the demands faced by workers (79, 80). These four items help identify challenges such as whether workloads are unevenly distributed or workers feel they are falling behind and how well they can manage their time. Through rigorous quantification of these dimensions, the tool enables a clear assessment of how these job demands contribute to overall stress and burnout levels. In this study, the scale has Cronbach's alpha was 0.84.

Perceived stress scale

A standardized PSS questionnaire was applied to measure the perceived work stress (72). It is a brief and simple tool with significant validity and reliability to measure the degree to which the conditions in one's life are perceived as stressful for the genesis of illness and behavioral disorders. The 14 items on the scale range from never = 0, almost never = 1, sometimes = 2, fairly often = 3, and very often = 4 were selected based on their relevance to the specific objective of this study. To achieve the tool's scores, the scores of seven positive items (four, five, six, seven, nine, ten, and thirteen) are reversed, such that 0 = 4, 1 = 3, 2 = 2, and so on, and then the 14 items are added up (72). This instrument is customized and tailored in Ethiopian cultures and used in various studies, and it was confirmed that the tool is reliable (82, 83). Moreover, the reliability (Chronbach's alpha) of PSS in this study was 0.77.

Job satisfaction

Job satisfaction was evaluated using a 10-item generic work satisfaction scale developed by MacDonald and MacIntyre (68). This scale, validated in Canada across various occupations and organizations, has proven to provide reliable and valid scores (68). In Ethiopia, the scale has been tailored into local languages and applied to diverse working populations, including school teachers, where it has also demonstrated excellent reliability (80, 84). The internal reliability of 10 items of job satisfaction scale has a Chronbach's alpha of 0.90.

Work–family conflicts scale

Work–family conflicts (WFCs) are key factors in occupational health, linked to reduced organizational commitment, poor job performance, and various negative outcomes like burnout, depression, and sleep issues. In this study, WFCs were evaluated using a 10-item self-report questionnaire that includes two scales: work–family conflict and family–work conflict. These scales are widely recognized for their reliability and validity across different professions and countries (70). In this study, a pretest was done to ensure clarity and cultural relevance of items for sensitivity to Ethiopian norms regarding work and family roles, and feedback from local educators helped modify language for better relevance. In addition, the internal reliability of Work–family conflicts 10 items has Chronbach's alpha of 0.86.

Working conditions scale

Working conditions scale (WCS) was adapted from previous literature to assess the conduciveness of class room conditions and school facilities (staff rooms, recreation centers, toilets, internet access, etc.), collegial relationships among teachers, and the number of kids in a class room, and dichotomized by their mean value (71). In the present study, the scales Cronbach's alpha was $\alpha = 0.89$.

Turnover intention scale

Turnover intention was (TIS) assessed using six items on a five-point Likert scale, indicating a desire to leave the job (73). In the current study, the scales Cronbach's alpha reliability ($\alpha = 0.907$) demonstrating high internal consistency.

Social support questionnaire

The three-item Oslo Social Support Questionnaire evaluates psychosocial resources within social networks. It's a brief, cost-effective tool used in various large-scale surveys, including the European Opinion Research Group (2003), Ireland's National Lifestyle Survey (2007), the European Kid Screen Study, and the Outcome of Depression European Network study (69). This instrument has been tailored and utilized in several studies conducted in Ethiopia, which confirm that the tool is reliable (85–87). In this study social support three items scale has a Chronbach's alpha values of 0.86.

The final versions of the questionnaire comprise four sections. The first section comprises socio-demographic characteristics, which assess information on age, sex, educational level, work experience, having children, monthly salary, working hour per day, and types of kindergarten. The second category encompasses questions to assess information on work-related burnout. The third part of the questionnaires includes behavioral factors like cigarette smoking, khat chewing, alcohol drinking, medication use for stress relevance, work misconduct issues, and physical exercise, and the fourth question assessed different factors in the workplace, such as work–family conflicts, job satisfaction, perceived stress, social support, working conditions, and the Copenhagen psychosocial questionnaire (62).

Data quality control

To ensure consistency, the survey instrument was first developed in English, translated into Amharic, the native tongue, and then back to English by language specialists. The questionnaires were pre-tested and structured. The pre-test was done on 5% of the total sample that was excluded in the final analysis. The reliability test from Chronbach's alpha was measured and calculated. The data were collected by two BSc-level environmental health professionals. One supervisor was recruited for continuous supervision of the data collectors. Data collectors and a supervisor received 2 days of training covering the goals of the study, data collection methods, and ethical guidelines. Every day, the lead investigator and supervisor reviewed the gathered data to ensure it was accurate, clear, and comprehensive, and any necessary changes were made immediately. Data cleansing and cross-checking were completed prior to analysis.

Data management and analysis

The data were imported into Epi-data version 4.6, exported, and cleaned before being analyzed using SPSS version 26 after accuracy tests. Cross-tabulations were conducted to evaluate the link between the outcome variable (WRB) and its associated factors, and frequency tables, and percentages were utilized to provide descriptive statistics. To handle confounding, we apply random sampling and multivariable regression analysis, and validated instruments were used to collect comprehensive data within a conceptual framework. The variables' multicollinearity, outliers, and normality were assessed before the bivariable and multivariable binary logistic regression analyzes were performed. The variance inflation factor was used to verify the multicollinearity assumption ($VIF < 2$). In the bivariable logistic regression analysis, variables demonstrating statistically significant correlations with the dependent variable at p-values < 0.2 were retained for further analysis. Using this p-value threshold is appropriate for preliminary screening to identify potential associations between the independent variables and the outcome of interest. This approach is valuable to avoid missing potentially important variables that could have significant implications for public health or policy. This higher p-value threshold helps ensure that potentially significant associations are not overlooked during this initial analysis phase. The model's fitness was checked by the Hosmer and Lemeshow goodness-of-fit ($p = 0.802$), and the results showed that it was well fitted (p value > 0.05). Finally, factors with a p-value < 0.05 were found using a multivariable binary logistic regression model. A 95% confidence interval-based adjusted odds ratio (AOR) was used to show the degree of relationship.

Results

Socio-demographic characteristics of participants

From a total of 422 selected KG teachers, 401 of them participated, with a response rate of 95%. The mean (±SD) age of the study participants was 28.5 (±5.8). The majority of the participants were female [391 (97.5%)], 240 (59.9%) were married, and 213 (53.1%) had at least one child. Regarding their educational level, 184 (45.9%) of them were diploma holders, and 87 (21.7%) of them had certificates. 174 (43.4%) of the participants had less than 5 years of teaching experience; 109 (27.2%) of them had less than a 3,000 Ethiopian birr (ETB) monthly salary; and the majority of 261 (65.1%) of them were taking early childhood care trainings (Table 1).

TABLE 1 Socio-demographic characteristics of kindergarten teachers in Dessie City, Northeast Ethiopia, 2024 ($n = 401$).

Variables	Frequency (n)	Percent (%)
Sex		
Male	10	2.5
Female	391	97.5
Age		
21–29	171	42.6
30–38	102	25.4
39–47	80	20.0
≥48	48	12.0
Religion		
Orthodox	239	59.6
Muslim	145	36.2
Protestant	17	4.2
Marital status		
Single	131	32.7
Married	240	59.9
Divorced	25	6.2
Widowed	5	1.2
Having children		
No children	188	46.9
At least one children	213	53.1
Number of children		
1–2	152	37.9
3–6	61	15.2
Educational level		
Certificate	87	21.7
Grade 11 and 12 complete	67	16.7
Diploma	184	45.9
Bachelor's Degree	63	15.7
Work-experience in years		
<5	174	43.4
5–10	113	28.2
11–15	61	15.2
≥16	53	13.2
Weekly class schedule		
1–4	42	10.5
5–6	359	89.5
Daily working hour		
≤8	369	92.0
>8	32	8.0
Monthly salary in Ethiopian birr		
1,609–3,000	109	27.2
3,100–4,000	98	24.4

(Continued)

TABLE 1 (Continued)

Variables	Frequency (*n*)	Percent (%)
4,001–5,100	95	23.7
5,101–10,000	99	24.7
Kindergarten type		
Public	185	46.1
Private	216	53.9
Taking early childhood care training		
Yes	261	65.1
No	140	34.9

Behavioral characteristics of participants

Of the participants, only 25 (6.2%) were alcohol drinkers, and none of them were smokers. The majority of participants were doing physical exercise for at least 2 days per week for 30 min. Among the study participants, 99 (24.7%) have work misconduct issues that may probably cause burnout in their professions. Only 57 (14.2%) of them used medication for stress related to their work (Table 2).

Work-related characteristics of kindergarten teachers in Dessie City

In terms of work-related characteristics, 190 (47.4%) had high job demands, and only 38 (9.5%) had low social support. Among them, 199 (49.6%) of them have work–family conflicts, and 220 (54.9%) of them work in poor working conditions. In terms of their role, 139 (34.7%) of them have poor role clarity, 190 (47.4%) were stressed, and 217 (54.1%) have a high turnover intention of leaving their job (Table 3).

Prevalence of work-related burnout

The total prevalence of WRB in the last 6 months was 39.7% ($n = 159$) [95% CI (34.8, 44.6)]. Table 4 displays the findings of all seven WRB items, together with the corresponding frequency and mean scores.

Factors associated with work-related burnout

In the bi-variable logistic regression analysis, educational level, kindergarten type, participant age, job demand, working condition, perceived stress, job satisfaction, work–family conflicts, work misconduct issue, work experience, and taking early childhood care training were factors associated with WRB ($p < 0.2$). Finally, after adjusting for confounding variables in the multivariable binary logistic regression analysis, only kindergarten type, job satisfaction, working conditions, perceived stress, and work–family conflicts were discovered to be statistically significant variables associated with WRB (p-value < 0.05).

TABLE 2 Behavioral characteristics of kindergarten teachers working in Dessie City, Northeast Ethiopia, 2024 ($n = 401$).

Variables	Frequency (*n*)	Percent (%)
Alcohol drinking		
Yes	25	6.2
No	376	93.8
Khat chewing		
Yes	4	1.0
No	397	99.0
Medication use		
Yes	57	14.2
No	344	85.8
Doing physical exercise		
Yes	263	65.6
No	138	34.4
Having work misconduct issue		
Yes	99	24.7
No	302	75.3

As a result, KG teachers who reported high work–family conflicts had a 2.8 times higher chance of developing WRB than those with low work–family conflicts [AOR: 2.81; 95% CI (1.77, 4.45)]. KG teachers who were not satisfied with their jobs showed a 1.8-fold increased risk of WRB compared to those satisfied with their job [AOR: 1.75, 95% CI (1.10, 2.79)]. Stressed KG teachers were 1.9 times more likely to suffer WRB than those who are not stressed [AOR: 1.91; 95% CI (1.21, 3.01)]. Compared to their peers', those who work under poor working conditions had a 1.6 times higher risk of experiencing work-related burnout [AOR: 1.63; 95% CI (1.03, 2.56)]. Teachers working in public KG schools had a 1.7-fold increased risk of getting WRB than their counterparts' [AOR: 1.66; 95% CI (1.02, 2.68)] (Table 5).

Discussion

This study aimed to investigating the prevalence of WRB and its associated factors among kindergarten teachers. As a result, the total prevalence of WRB in the last 6 months was 39.7% [95% CI (34.8, 44.6)]. This indicates that burnout level among kindergarten teachers in Dessie City, Ethiopia, was high. In addition, work–family conflicts, working conditions, perceived stress, job satisfaction, and kindergarten type were factors significantly related to WRB. This study's findings are grounded in structural theory that illustrates how an organization's structure and culture impact employee well-being, and an unmanaged stress level causes burnout (58). In addition, the findings of this research are supported by demand-resource theory that posits burnout occurs when the demands of work, such as high perceived stress and poor working conditions, lead to chronic fatigue and ultimately burnout (60). Moreover, the results of this study are grounded in Social Exchange Theory, which explains burnout arises in the social and interpersonal context of the work organization when there is a lack of reciprocity at the organizational level or when employees' expectations regarding the nature of their exchange with

TABLE 3 Work-related characteristics of kindergarten teachers in Dessie City, Northeast Ethiopia, 2024 (*n* = 401).

Variable	Frequency (*n*)	Percent (%)
Job demand		
High	190	47.4
Low	211	52.6
Job recognition		
High	272	67.8
Low	129	32.2
Role clarity		
Good	262	65.3
Poor	139	34.7
Social support		
Low	38	9.5
Moderate	175	43.6
High	188	46.9
Work–family conflicts		
High	199	49.6
Low	202	50.4
Working conditions		
Good	181	45.1
Poor	220	54.9
Job Satisfaction		
Satisfied	164	40.9
Dissatisfied	237	59.1
Perceived Stress		
Stressed	190	47.4
Not Stressed	211	52.6
Turnover intention		
High	217	54.1
Low	184	45.9

the organization are not met, which creates work–family conflicts and makes them dissatisfied with their job (61).

The prevalence of WRB in this finding was almost similar to studies conducted among school teachers in Brazil (36.7%) (88), Nigeria (36%) (89), and Ethiopia (37.4%) (79). This resemblance could be explained by the long working hours and intense nature of the teaching profession, which contribute to chronic stress and increases the risk of burnout (46). However, the current study finding is higher as compared with kindergarten teachers in Germany (24.4%) (28), and school teachers in Iraq (24.5%) (90), Namibia (28.8%) (91), and Tunisia (27.4%) (35). The possible reason for this might be kindergarten teachers need to put in sufficient physical and psychological effort to accomplish their work, but KG teachers in Dessie City gain significantly lower recognition, a low salary, and low social-economic status, plus their work nature leads to high burnout. Besides, their salary is insufficient to administer their lives; they have a dual role as caregiver and educator, which may exhaust their energy and cause burnout (92). In addition, their work is demanding due to the nature of the children, who are immature, and they are always in touch with children, and their role is not limited to teaching and conveying information to children but instead has a role with various aspects and characteristics as a role in developing the educational process. Furthermore, kindergarten teachers having a long working hour, high intensity, and wide range of emotional interactions (93) with their students causes them stress, fatigue, emotional exhaustion, and more burnout, which decreases teaching performance and leads to less interaction with students in their classrooms (14). Other possible reasons may be variations in study settings, tool differences, and methods of data collection. In contrast, this study indicated a decreased prevalence of WRB compared to studies in China (27). This may be due to cultural differences, variations in measurement tools, and the specific contexts in which the teachers work.

This study found that work–family conflicts were associated with WRB. This finding is supported by studies in India (94), Indonesia (52), Portugal (53), Hungary (54), Malaysia (55), and China (34). This might be because the majority, or all, of the KG teachers were female teachers. They performed dual roles, managing both their job responsibilities and family care, and experienced strain from work

TABLE 4 Response categories and scoring system of work-related burnout among kindergarten teachers in Dessie City, Northeast Ethiopia, 2024 (*n* = 401).

Work-related burnout items (Chrombach's alpha = 0.82)	Response category and scoring system					
	Never scoring	Seldom scoring	Some times scoring	Often scoring	Always scoring	Score mean (SD)
	0%	25%	50%	75%	100%	
1. Is your work emotionally exhausting?	11.2	29.9	29.9	11.5	17.5	48.50 (31.24)
2. Do you feel burnout because of your work?	16.5	35.4	26.2	12.0	10.0	40.90 (29.60)
3. Does your work frustrate you?	31.4	30.9	24.2	8.7	4.7	31.11 (28.23)
4. Do you feel worn out at the end of the working day?	13.5	29.9	28.2	13.2	15.2	46.70 (31.27)
5. Are you exhausted in the morning at the thought of another day at work?	34.2	27.4	24.2	11.2	3.0	30.36 (28.05)
6. Do you feel that every working hour is tiring for you?	20.4	31.7	27.2	14.0	6.7	38.72 (28.97)
7. Do you have enough energy for family and friends during leisure time? (Reversed scoring)	1.7	7.2	28.4	31.4	31.2	70.76 (25.20)
Total average score						43.86
						28.94

TABLE 5 **Bi-variable and multivariable binary logistic regression analysis of associated factors with work-related burnout among kindergarten teachers in Dessie City, Northeast Ethiopia, 2024 (n = 401).**

Variables (N = 422)	Work-related burnout		COR with 95% CI	AOR with 95% CI
	Yes	No		
Age				
21–29	78	93	1.68 (0.86, 3.28)	1.46 (0.68, 3.16)
30–38	46	56	1.64 (0.80, 3.36)	1.96 (0.83, 4.60)
39–47	19	61	0.62 (0.28, 1.37)	0.57 (0.24, 1.39)
≥48	16	32	1	1
Work experience in year				
<5	86	88	2.26 (1.17, 4.36)	1.84 (0.88, 3.88)
5–10	43	70	1.42 (0.71, 2.86)	0.93 (0.42, 2.06)
11–15	14	47	0.69 (0.30, 1.59)	0.53 (0.21, 1.36)
≥16	16	37	1	1
Kindergarten type				
Public	88	97	1.85 (1.24, 2.78)	1.66 (1.02, 2.68)*
Private	71	145	1	1
Work misconduct issue				
Yes	46	53	1.45 (0.92, 2.30)	1.47 (0.85, 2.54)
No	113	189	1	1
Early childhood care training				
Yes	97	164	1	1
No	62	78	1.34 (0.89, 2.04)	1.29 (0.80, 2.09)
Job satisfaction				
Dissatisfied	107	130	1.77 (1.17, 2.69)	1.75 (1.10, 2.79)*
Satisfied	52	112	1	1
Perceived stress				
Stressed	90	100	1.85 (1.24, 2.78)	1.91 (1.21, 3.01)*
Not stressed	69	142	1	1
Job demand				
High	83	107	1.38 (0.92, 2.06)	1.44 (0.92, 2.27)
Low	76	135	1	1
Work–family conflicts				
High	100	99	2.45 (1.62, 3.69)	2.81 (1.77, 4.45)**
Low	59	143	1	1
Working conditions				
Poor	98	122	1.58 (1.05, 2.37)	1.63 (1.03, 2.56)*
Good	61	120	1	1

Crude odds ratio (COR); confidence interval (CI); adjusted odds ratio (AOR); reference category (1); **statistically significant at $p < 0.001$; *statistically significant at $p < 0.05$; Hosmer and Lemeshow test = 0.802; it showed that the model fitted well.

demands that interfered with their home responsibilities. This ultimately led to the spillover of high work demands into their home life, causing depletion of energy and contributing to job burnout (55). WFCs affects work engagement by inducing stress, tension, sleep disturbances, and other adverse psychological and physical symptoms that trigger the energy exhaustion of KG teachers and lead to burnout (95, 96). If kindergarten teachers are unable to fulfill the expectations of their families and employers, this condition may lead to disputes and disagreements that can subsequently result in mental pressure and experiences of burnout (97). Another possible reason is the prolonged working hours of KG teachers, which are 9 h and sometimes 10 h in school per day. This extensive time spent at work can lead to neglecting household responsibilities and family care, exacerbating WFCs. When challenges at home and in their social lives hinder teachers' ability to fulfill work obligations, it creates family–work conflict, resulting in time pressures, reduced flexibility, fatigue, and energy depletion. Consequently, teachers may devote less time to their families, which can intensify WFCs and ultimately lead to burnout (98). Additionally, the cost of living rising in Ethiopia places further burdens on teachers, compelling them to manage their professional, social, and familial responsibilities simultaneously, thus contributing to WFCs and increasing the risk of stress and burnout.

In this study, job dissatisfaction was associated with WRB. This finding is corroborated in studies conducted in Malaysia (99), Norway (100), and Italy (30). This is due to the fact that job dissatisfaction is associated with high levels of burnout (101). In addition, job satisfaction, which is the overall feeling about one's job or career, is used as a buffer against burnout, and their job satisfaction has implications for student learning in that a satisfied teacher may provide better quality or more consistent instruction to his or her students. Furthermore, teachers with low salaries and poor working conditions may lose their emotional resources and job satisfaction, leading to energy depletion and increased susceptibility to burnout. Conversely, when teachers experience a decline in job satisfaction, it can heighten feelings of cynicism and reduce their sense of personal accomplishment, which may further contribute to burnout (102).

In addition, the findings of this study highlight that working conditions are statistically associated with WRB. This finding is concurrent with findings in Tanzania (103), Iraq (90), Sweden (51), and Brazil (104). Possible reasons for this correlation might be the presence of factors such as inadequate school facilities, work overload, a high number of students per class, and poor staff relations. KG teachers working under these challenging conditions may feel pressured and stressed, consequently increasing their likelihood of developing WRB. Besides, a highly demanding work atmosphere can lead to strain reactions in teachers and impair their work performance, making it one of the key factors related to burnout. Moreover, KG teachers face numerous challenges in the development of society and education, which contribute to increased workloads. These challenges include curriculum adaptation, the use of diverse educational tools, and effective classroom management. When employees are unable to cope with these demanding working conditions, they may feel restless, fatigued, exhausted, and ultimately experience burnout. Furthermore, the short breaks between classes contribute to the exhaustion felt by KG teachers, as they are under significant pressure from parents and their expectations. This pressure can lead teachers to experience detachment, emotional exhaustion, and cynicism (34). Another likely reason is that KG teachers' work under conditions that require them to take charge of children's safety and care, which can lead to high job stress. This stress often contributes to negative emotional issues such as despair, anxiety, and burnout (105).

Moreover, in this study, perceived workplace stress is associated with burnout. This finding is in agreement with the findings in Japan (106), China (27, 107), and the South Central United States (108). One possible explanation is that educators who experience high levels of stress are having a high chance of developing WRB (109). The challenging and unique nature of their work, including managing crying children, dressing and undressing them, overseeing lunchtime and clean-up, monitoring nap times, and continuous exposure to germs and illness, contributes to this stress. Additionally, the lack of enough breaks during their workday and unclear delineation of work duties can lead to frustration and heightened stress, ultimately resulting in burnout (110). Another likely explanation is that they may experience heightened perceived stress due to a demanding workload, the number of kids per classroom, lack of resources, poor pay, low job status, numerous classroom responsibilities, curriculum demands, testing, performance evaluations, disruptive students, and the burden of providing direct care for preschool children and meeting their needs (59). These factors are significant contributors to stress and ultimately lead to burnout (111, 112). Moreover, the peculiar nature of their work, such as providing verbal and visual support, offering repeated explanations, assisting with the development of social skills, orienting children in space and time, resolving conflicts, facilitating communication and social relationships, supervising rest periods, and preparing teaching aids and materials can contribute to stress that depletes their energy and ultimately leads to burnout (113). Lastly, kindergarten teachers must develop creative ways to make their classes engaging and visually appealing. Some of these strategies include role plays, singing, dancing, interactive games, creating an interactive stage, drawing, and offering prize incentives. However, while these activities can enhance the learning environment, they can also become sources of stress and exhaustion for teachers. If they are unable to manage these demands, it may ultimately lead to burnout (114).

Lastly, in this study, public kindergarten teachers had more burnout as compared to their private counterparts. This finding is supported by a study in China (27). The possible explanations might be that private kindergartens are often newer, have smaller class sizes, and their head instructors have a high chance of receiving training in early childhood care compared to public kindergartens (115). Additionally, kids enrolled in private kindergartens typically come from wealthier families and tend to score higher on school readiness assessments. This readiness is essential for facilitating the teaching and learning process, which may reduce the burden on teachers (115). On the contrary, public kindergarten teachers were less likely to get the chance of receiving early childhood care training, which may result in lower scores on emotional support and behavior management in their classrooms (108). This is evidenced by our observations during data collection, which highlighted that in a public kindergarten, there were a large number of children in one class, with insufficient sleeping space during nap time. This overcrowding can lead to behavioral disturbances and create challenges for the teachers. Moreover, lack of facilities such as separate mail areas, hand washing stations, and adequate playgrounds with necessary materials increased the burden on teachers to address these gaps, ultimately leading to heightened stress and difficulties in supporting the children effectively. On top of that, there are differences in the required qualifications and work environments between public and private kindergartens. Educators in private schools have autonomy, administrative support, and minimal bureaucratic layers; they tend to be more professionally certified and may be less likely to experience burnout (116). Conversely, while teachers in public school are regarded as public servants and are guaranteed jobs until retirement, along with a pension after retiring, they face significant challenges. These include high workloads, insufficient salaries, low recognition and promotion opportunities, limited resources that hinder the teaching-learning process, and unsatisfactory working conditions. These factors can intensify the level of burnout among public kindergarten teachers (117).

Strengths and limitations of the study

This study used a standardized and reliable tool with a sufficient sample size from several kindergarten schools, including both public and private, and can be used as baseline information for programmers and other researchers. In addition, this study may contribute to the growing corpus of research on WRB and its predictors in this population. However, the study was based on a cross-sectional study design, which hinders the temporal relationship between WRB and factors affecting its development. Moreover, since the source data were self-reported from prior experiences of kindergarten teachers, recall and social desirability bias may underestimate or overestimate the level of burnout. To reduce this bias, we restrict the data to recent experiences only. The study focuses on kindergarten teachers in a specific city, which might overlook factors that could be relevant in other educational levels or geographical areas within the country. To demonstrate the association between different workplace aspects and WRB, future research must take into account a range of workplaces. With the exception of these limitations, we think this study provides trustworthy and robust data to address kindergarten teachers' work-related burnout in Dessie, Ethiopia.

Implications of the study

The study's findings are crucial in shaping educational policies and support programs tailored to Ethiopian kindergarten teachers, considering the unique socio-cultural and economic factors in the country. The study advocates for policies that prioritize teacher well-being as a key component of educational quality. This could include regular mental health assessments and support integrated into the national teacher support framework. The findings suggest implementing professional development programs focused on stress management, work-life balance, working conditions, job satisfaction, and coping strategies designed specifically for kindergarten teachers. These could be integrated into existing teacher training or offered as ongoing professional development. In addition, the research recommends creating localized teacher support networks such as strategies for improving teacher-student ratios, financial incentives, stress management workshops allowing teachers to share experiences, seek peer support, and access resources to manage burnout. These networks could be supported by local educational authorities. Moreover, this study suggests long-term policy recommendations, such that the study calls for the establishment of long-term monitoring and evaluation mechanisms to continuously assess teacher burnout. This could lead to a national framework for teacher well-being aligned with Ethiopia's broader educational goals.

Conclusion

According to this study, a significant number of kindergarten teachers were affected by burnout due to their working nature. KG teachers' WRB was found to be predicted by factors such as work–family conflicts, job satisfaction, perceived stress, kindergarten type, and working conditions. Interventions focused at reducing work–family conflicts, increasing job satisfaction, and improving the school environment are needed to address this critical issue and support teachers' well-being for the benefit of young students and the wider educational landscape.

Data availability statement

The original data is provided in the article, and further inquiries are directed to the corresponding author upon request.

Ethics statement

This study was conducted respecting the Helsinki declaration of research ethics for studies with human participants, and the ethical soundness of the research was approved by the Institutional Review Committee of the Department of Environmental and Occupational Health and Safety, University of Gondar, with the approval number EOHS/105/2024. The patients/participants provided their written informed consent to participate in this study.

Author contributions

AB: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Resources, Software, Validation, Visualization, Writing – original draft, Writing – review & editing. GZ: Conceptualization, Methodology, Validation, Visualization, Writing – review & editing. AT: Conceptualization, Methodology, Validation, Visualization, Writing – review & editing. AY: Conceptualization, Methodology, Validation, Visualization, Writing – review & editing. AA: Conceptualization, Methodology, Validation, Visualization, Writing – review & editing. GA: Conceptualization, Data curation, Methodology, Project administration, Resources, Visualization, Writing – review & editing.

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Conflict of interest

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

Publisher's note

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EDITED BY
Hubert Amu,
University of Health and Allied Sciences, Ghana

REVIEWED BY
Filippo Quattrone,
Sant'Anna School of Advanced Studies, Italy

*CORRESPONDENCE
Wiebe Külper-Schiek
✉ KuelperW@rki.de

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Evaluation of National Immunization Technical Advisory Groups (NITAGs) of middle-income countries in the WHO European Region; a synopsis

Wiebe Külper-Schiek[1]*, Liudmila Mosina[2], Lisa A. Jacques-Carroll[2], Annika Falman[1], Thomas Harder[1], Eduard Kakarriqi[3], Iria Preza[4], Arman Badalyan[5], Gayane Sahakyan[6], Oxana Romanova[7], Veronika Shimanovich[8], Sanjin Musa[9], Dinagul Bayesheva[10], Nurshay Azimbayeva[11], Zuridin Nurmatov[12], Vera Toigombaeva[13], Ninel Revenco[14], Veaceslav Gutu[15], Ljiljana Markovic-Denic[16], Branka Bonaci-Nikolic[16,17], Dilorom Tursunova[18], Nigora Tadzhiyeva[19], Ole Wichmann[1] and Siddhartha Sankar Datta[2]

[1]Infectious Disease Epidemiology, Robert Koch Institute, Berlin, Germany, [2]WHO Regional Office for Europe, Vaccine-Preventable Diseases & Immunization, Copenhagen, Denmark, [3]Epidemiology and Control of Infectious Diseases Department, Institute of Public Health of Albania, Tirana, Albania, [4]WHO Country Office for Albania, Vaccine-Preventable Diseases & Immunization, Tirana, Albania, [5]Department of Epidemiology, Yerevan State Medical University, Yerevan, Armenia, [6]National Center of Disease Control and Prevention, Ministry of Health of Armenia, Yerevan, Armenia, [7]Department of Pediatric Infectious Diseases, Belarus State Medical University, Minsk, Belarus, [8]Laboratory of Quality Control of Immunobiological Medicines, Republican Research and Practical Center for Epidemiology and Microbiology, Minsk, Belarus, [9]Department of Infectious Disease Epidemiology, Institute for Public Health of the Federation of Bosnia and Herzegovina, Sarajevo, Bosnia and Herzegovina, [10]Department of Children's Infectious Diseases, Astana Medical University, Astana, Kazakhstan, [11]Vaccine Preventable Disease Control Unit of the Sanitary - Epidemiology Control Committee, Ministry of Health of the Republic of Kazakhstan, Astana, Kazakhstan, [12]National Institute of Public Health, Ministry of Health of the Kyrgyz Republic, Bishkek, Kyrgyzstan, [13]Department of General and Clinical Epidemiology, Kyrgyz State Medical Academy, Bishkek, Kyrgyzstan, [14]Department of Pediatrics, State University of Medicine and Pharmaceutics "Nicolae Testemiţanu", Chisinau, Moldova, [15]Department of Vaccine-Preventable Diseases, National Agency for Public Health of Moldova, Chisinau, Moldova, [16]Faculty of Medicine, Institute for Epidemiology University of Belgrade, Belgrade, Serbia, [17]Department of Allergy and Immunology, Faculty of Medicine, University of Belgrade and Clinic of Allergy and Immunology/Clinical Center of Serbia, Belgrade, Serbia, [18]Committee for Sanitary-Epidemiological Welfare and Public Health of Republic of Uzbekistan, Immunization and Logistics, Tashkent, Uzbekistan, [19]Department of Infectious Diseases, Tashkent Medical Academy, Tashkent, Uzbekistan

Introduction: A National Immunization Technical Advisory Group (NITAG) provides independent guidance to Ministries of Health (MoH) and policymakers, enabling them to make informed decisions on national immunization policies and practices. As of 2022, 50 of the 53 countries in the World Health Organization (WHO) European Region (the Region) had established a NITAG, with 58% of all NITAGs and 66% of those in middle-income countries (MICs) in the Region meeting all six WHO process indicators of NITAG functionality. However, many newly established NITAGs in MICs in the Region experience challenges in terms of their functioning, structure, and outputs.

Methods: To address these challenges and achieve the goal of evidence-informed decision making on immunizations, the WHO Regional Office for Europe and the Robert Koch Institute (RKI) implemented a project to strengthen the functioning of MIC NITAGs of the Region through comprehensive evaluations of nine NITAGs and development and implementation of improvement plans.

Results: All evaluated NITAGs are formally established and complete the most important aspects of NITAG functioning. The main challenge for all NITAGs is the lack of a well-staffed Secretariat to establish annual workplans and develop NITAG recommendations following a standardized process.

Discussion: The evaluation identified NITAGs' strengths and challenges. Some challenges have been addressed through improvement plan implementation. WHO and RKI will continue to evaluate NITAGs and support development and implementation of improvement plans. WHO and NITAG partners will continue to provide training on the standardized recommendation-making process and advocate increased MoH support to NITAGs, including dedicated Secretariat staff.

KEYWORDS

immunization, evidence-based decision making, middle-income countries, National Immunization Technical Advisory Group (NITAG), evaluation, immunization policy, vaccination, World Health Organization

1 Introduction

A National Immunization Technical Advisory Group (NITAG) is composed of multi-disciplinary experts who provide scientific evidence and support to Ministries of Health (MoH) and governments in making evidence-informed decisions related to immunization policies and practices (1, 2). The NITAG's role is to strengthen country ownership and public confidence in the national immunization programme by developing national recommendations that are based on the best available evidence using a transparent and systematic process to increase the credibility of MoH or government decisions and build the resilience of National Immunization Programmes (NIPs) (3, 4). In recent years, many low and middle-income countries (LMICs) have followed the lead of high-income countries by establishing NITAGs; the Global Vaccine Action Plan 2011–2020 called on all countries to establish or have access to a NITAG by 2020 (5).

As of 2022, 50 out of 53 countries in the World Health Organization (WHO) European Region (the Region), including all 18 MICs, reported having a NITAG in place through the WHO/UNICEF Joint Reporting Form (JRF) (6). However, performance varies widely; in 2022, 58% of all NITAGs and 66% of NITAGs in MICs reported meeting all six process indicators of NITAG functionality. The main challenges in meeting the six indicators were in collecting a declaration of interest from all NITAG members, a lack of data on the number of meetings in the reporting year, and insufficient representation of the five required disciplines by NITAG members.

Evaluating NITAGs' structure, functioning and work processes helps NITAGs identify areas for improvement. Such evaluations have been conducted in the past by WHO and NITAG partners (e.g., the Supporting Independent Immunization and Vaccine Advisory Committees [SIVAC] initiative that conducted evaluations of the NITAG of Armenia in 2015, and of the NITAG of the Republic of Moldova in 2016) (7–11). Evaluation reports were provided to the team by the NITAGs. Additionally, in 2016, WHO conducted a survey to evaluate NITAGs from MICs. This survey revealed that the composition and function of some NITAGs were still not in line with WHO recommendations and most NITAGs of MICs did not have a systematic recommendation-making process.

Based on the findings from the SIVAC evaluations and the WHO survey, NITAG strengthening activities implemented in the Region from 2017 to 2019 focused on increasing NITAGs' functionality and capacity to develop systematic evidence-based recommendations (8). The Evidence to Recommendation (EtR) process is used by many long-functioning NITAGs such as the United States Advisory Committee on Immunization Practices (ACIP), Germany's Standing Committee on Vaccination (STIKO), and WHO's Strategic Advisory Group of Experts on Immunization (SAGE) and includes a process of systematic collection, quality-assessment, and synthesis of evidence, which allows for transparent communication of the evidence that leads to a recommendation (12–14). To assess the status of NITAG functionality in the Region after the implementation of strengthening activities and to gain an understanding of the remaining challenges, the Regional Office and Robert Koch Institute (RKI) initiated a joint project in 2020, the *EURO NITAGs Project,*[1] with financial support from the German MoH. The project aims to conduct in-depth evaluations of NITAGs in 16 MICs in the Region, support NITAGs in developing and implementing improvement plans to address identified challenges and increase NITAGs' capacity to develop evidence-informed immunization policy recommendations.

This article outlines the project support provided, the evaluation process, and a concise summary of major evaluation results.

2 Methods

2.1 Evaluation tool

The evaluations were conducted using a detailed *Evaluation Tool for NITAGs* (15) (referred to hereafter as "the questionnaire")

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[1] In 2023, the EURO NITAGs Project continued within the SENSE-project (Strengthening National Immunization Technical Advisory Groups and their Evidence-based Decision-making in the WHO European Region and globally; https://ghpp.de/en/projects/sense/).

developed specifically for this project. The questionnaire was developed by reviewing the structure, questions and answer options of existing evaluation tools (e.g., SIVAC evaluation tool, WHO NITAG Simplified Evaluation Tool) (16, 17). While the structure of the questionnaire is in line with those of existing tools, we rephrased, combined and added more detailed questions on specific aspects to allow the study team to get an in-depth understanding of the functioning of the MIC-NITAGs in the Region and identify specific strengths and challenges. The questionnaire includes questions covering three evaluation areas: (1) NITAG functionality, which includes the formal establishment of the NITAG, its membership and composition, available resources, funding, and independence; (2) Quality and results of the NITAG's work processes including the preparation and conduct of meetings, and the recommendation-making process; and (3) NITAG's integration into decision-making processes, including collaboration between the NITAG and MoH and other immunization stakeholders and the NITAG's public visibility. NITAGs complete the questionnaire, self-assess their performance in each area, and summarize their main strengths and challenges. To ensure clarity of the questions and usability of the tool, the questionnaire was piloted in two countries (in-country evaluation in Belarus and self-evaluation in Albania) and revised based on the countries' feedback.

The final version of the questionnaire is published on the Regional Office website and includes an instruction guide and NITAG improvement plan template (15).

2.2 Evaluation methodology

The NITAG evaluations are conducted in four phases. During phase 1, the project team (Regional Office and RKI) conducts a briefing on the evaluation process with the NITAG Chair and Secretariat, obtain the NITAG's commitment to conduct the evaluation, and collect relevant documents such as meeting minutes, terms of reference (ToR), standard operating procedures (SOPs), and recently developed recommendations. During phase 2, the NITAG completes the questionnaire independently (self-evaluation) or with the project team's support (external evaluation). In phase 3, the project team reviews the completed questionnaire and relevant documents. Any unclear or inconsistent information from the questionnaire or shared documents is discussed with the NITAG Secretariat and/or Chair and misunderstandings of terminology and concepts are explained and clarified. Based on this discussion, the project team develops a detailed report for each NITAG including strengths and challenges identified by both parties and recommendations to overcome identified challenges. During phase 4, the NITAG, with the project team's support, develops an improvement plan based on the recommendations, including interventions and detailed activities for each area of improvement, persons responsible for each activity, NITAG partners to be involved, and an implementation timeframe.

Between 2020 and 2023, the project team conducted evaluations of nine NITAGs including Albania, Armenia, Belarus, the Federation of Bosnia and Herzegovina (Bosnia and Herzegovina), Kazakhstan, Kyrgyzstan, the Republic of Moldova, Serbia, and Uzbekistan. Evaluations of the remaining seven MIC NITAGs are scheduled for 2024–2025.

3 Results

3.1 Evaluation results

In the following section, the major results of the NITAG evaluations are presented. For the tabular presentation, the questions and sub-questions from the questionnaire were summarized and re-structured to provide overarching evaluation questions. Three assessment categories (fully yes, partially yes, and no) describe the NITAGs' functionality (Table 1), work processes, outputs, and integration into the policy process (Table 2) and are linked to the questions and key aspects indicated in the tables. Further details on the NITAG assessments are available as Supplementary material.

3.1.1 Formal establishment

All NITAGs were formally established as an advisory body through a MoH order. Most of the NITAGs ($n = 7$) have a document (e.g., ToR) that describes their functioning, however, two out of the seven do not address all relevant aspects that define the functioning of NITAGs in the ToR (see key aspects considered in Table 1).

3.1.2 Membership and composition

All NITAGs have core members representing experts from various disciplines to decide on final NITAG recommendations. However, three NITAGs include core members who work for the MoH or NIP and therefore are not independent experts. All NITAGs have an appointed Chair and, except for one NITAG, the role of the Chair is defined in the NITAG's ToR. All NITAGs have a Secretariat in place to provide technical support to the NITAG. However, none of the evaluated NITAG Secretariats are considered "fully functional" (see key aspects considered in Table 1). The major challenge is the absence of dedicated Secretariat staff who can provide technical support to the NITAG. For most NITAGs, National Public Health Institute (NPHI) officers or NIP staff conduct Secretariat work in addition to their routine responsibilities. For two NITAGs, the Secretary also serves as the NITAG Chair (Bosnia and Herzegovina) or as a core NITAG member (Republic of Moldova)[2]. Five NITAGs have established working groups (WGs) to prepare specific topics for NITAG discussions while only three of these have developed a WG ToR. Three NITAGs have not established WGs due to resource constraints (human and time) or a lack of experts willing to serve in WGs.

3.1.3 Resources and funding

Only one NITAG had secured sustainable funding to cover expenses for NITAG meetings including per diem for NITAG members.

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[2] As the aim of a Secretariat is to provide technical support to the NITAG by collecting and synthesizing evidence for NITAG recommendations, Secretariat staff is not fully independent and should not be involved in the NITAGs discussions and/or final NITAG recommendation-making.

TABLE 1 Description of the functionality of the evaluated NITAGs.*

	Establishment of NITAG		Membership and composition					Resources and funding		Independence
Questions on NITAG functionality	**Is the NITAG formally established as an advisory body?**	**Is the functioning of the NITAG clearly defined in a document?**	**Does the NITAG include voting (core) members that are independent from the MoH/NIP and represent most disciplines?**	**Has the NITAG designated a Chair with a defined role?**	**Does the NITAG have a fully functional Secretariat?**	**Does the NITAG include non-voting (non-core) members?**	**Does the NITAG establish working groups (WGs) for specific topics?**	**Does the NITAG have access to various databases and external experts' consultations?**	**Are NITAG activities sustainably funded/ financially supported?**	**Does the NITAG follow a policy on conflicts of interest (CoI) for core-members?**
Key aspects considered	Official establishment by MoH; availability of the document; the NITAG is an advisory body	Document available that describes the NITAG's functioning including all relevant aspects[†]	Core members cover majority of expertise[^]; no core members work in MoH/NIP	Chair in place; role of Chair defined	Secretariat includes ≥1 person with ≥50 FTE%; not part of NITAG core members; provides minimum basic technical support		≥1 WG currently or established in past; WG-ToR available; WG reports to Secretariat	Access to local/ regional/national data and scientific databases available; experts for NITAG consultation available (other than included in WG)	MoH provides funding for NITAG activities	Written declaration of interest; consequences of CoI pre-defined; external assessment of existing conflict (e.g., by Chair/ Secretariat/legal office)
Albania	FY	FY	FY	PY	PY	N	PY	FY	N	PY
Armenia		FY	FY	FY		FY	FY	FY	N	FY
Belarus		PY	PY				N	FY	N	PY
Federation of Bosnia and Herzegovina		PY	PY				N	PY	PY	N
Kazakhstan		FY	FY				N	FY	N	PY
Kyrgyzstan		FY	FY				FY	FY	N	PY
Republic of Moldova		FY	FY				PY	FY	N	PY
Serbia		N	FY			N	FY	FY	N	PY
Uzbekistan		N	PY			FY	PY	PY	N	N

*The assessment categories are defined by the project team based on the phase 3 review and discussion of evaluation tool responses.

[†] Relevant aspects that define the functioning of the NITAG include: activity planning procedures; minimum number of meetings per year; quorum for conducting a meeting/making a decision; type and number of members, roles, and length of mandate; policy on conflicts of interest; Secretariat role and functioning; procedures related to the circulation of background materials and meeting agenda.

[^] The NITAG should include representation of the following disciplines: pediatrics, public health experts, infectious diseases experts, epidemiology experts, immunology.

Key: FY, Fully Yes ("yes" to all aspects); PY, Partially Yes (not all aspects are answered by "yes"); N, No ("no" to all aspects); CoI, conflict of interest; FTE, full-time equivalent; NIP, National Immunization Programme; NITAG, National Immunization Technical Advisory Group; MoH, Ministry of Health; WG, working group. For additional details, see Supplementary material.

TABLE 2 Description of the work processes, outputs, and integration into the policy process of the evaluated NITAGs.*

	NITAG meetings			Development of NITAG recommendations				Integration into policy processes	
Questions on NITAG work processes and outputs and integration into policy processes	**Does the NITAG have an annual work plan?**	**Does the NITAG meet regularly and according to pre-defined meeting frequency?**	**Are NITAG meetings formally prepared and followed-up?**	**Does the NITAG develop recommendations using a standardized process?**	**Are NITAG recommendations shared with MoH?**	**Are the majority of NITAG recommendations accepted and implemented by MoH?**	**Are NITAG recommendations publically available?**	**Is the NITAG well recognized among stakeholders and the public and regularly consulted by MoH on immunization aspects?**	**Does the NITAG collaborate with relevant partners?**
Key aspects considered	Annual work plan developed; NITAG works according to the work plan	NITAG meets regularly (≥ 1 meeting/year); adherence to pre-defined meeting frequency	Meeting agenda developed and shared ≥2 weeks before the meeting; background document compiling collected evidence developed and shared ≥1 week before the meeting; minutes/reports prepared after the meeting	Recommendation process includes all relevant aspects[†]	Recommendations are shared with MoH (specific MoH person in charge); document includes recommendation and concise summary of evidence (e.g., policy report)	Majority of developed recommendations are accepted and implemented by MoH		Regular consultation by MoH; recognition among experts, stakeholders, MoH, public	Collaboration with partners/networks (e.g., other NITAGs, NITAG Network)
Albania	FY	PY	PY	PY	FY	FY	PY	PY	FY
Armenia	FY	FY	FY		PY		FY	PY	FY
Belarus	FY	PY	PY		PY		N	PY	PY
Federation of Bosnia and Herzegovina	N	FY			PY		FY	FY	N
Kazakhstan	FY				FY		PY	FY	PY
Kyrgyzstan	PY				PY		N	PY	FY
Republic of Moldova	FY				PY		PY	FY	N
Serbia	N				PY		FY	PY	FY
Uzbekistan	FY			N	PY		N	PY	N

*The assessment categories are defined by the project team based on the phase 3 review and discussion of evaluation tool responses.

[†] Relevant aspects that should be included in a systematic recommendation-making process are the following: (1) formulation of a policy question, (2) use of pre-specified criteria, (3) collection of evidence according to defined criteria, (4) assessment of the quality of evidence, (5) systematic synthesis of evidence.

Key: FY, Fully Yes ("yes" to all aspects); PY, Partially Yes (not all aspects are answered by "yes"); N, No ("no" to all aspects); MoH, Ministry of Health; NITAG, National Technical Immunization Advisory Group. For additional details, see Supplementary material.

3.1.4 Independence

Only one NITAG requests core members to declare their interests in writing and assesses declared interests externally (e.g., the NITAG Chair or Secretariat determines whether the declared interest could have any influence on the discussion topic), and has a pre-defined process for managing existing or perceived conflicts of interest (CoIs). Kyrgyzstan's NITAG includes all aspects in their ToR, but not all are implemented. Six NITAGs have a CoI policy that is either based on only oral declarations, or self-assessments of existing conflicts or does not pre-define how to manage identified conflicts. Two NITAGs have no CoI policy.

3.1.5 NITAG meetings

All but one NITAG aligns the discussion topics with the goals and targets of the NIP. Seven NITAGs develop an annual work plan that prioritizes topics throughout the year. The remaining NITAGs do not have an annual plan but define topics before meetings. All NITAGs meet regularly and provide background materials to members before the meeting. However, only one NITAG prepares background documents with a concise summary of the collected evidence, facilitating focused and effective deliberations. All NITAGs submit meeting minutes or reports to their MoHs.

3.1.6 Development of NITAG recommendations

Seven out of nine NITAGs have a pre-defined process to develop recommendations. However, none of the evaluated NITAGs implement all aspects of the EtR process (see explanation in Table 2) in their recommendation-making mechanisms. Most NITAGs do not develop structured policy questions, assess the quality of the collected evidence, and/or systematically synthesize the collected evidence. Reasons for not applying a systematic process were diverse, including a lack of human resources and time to conduct such a process or a lack of awareness of the importance of the process. NITAG recommendations are shared with the MoH mainly in the form of meeting minutes. A separate document (e.g., policy brief) that includes a concise summary of the evidence resulting in the NITAG recommendations is developed only by Kyrgyzstan's NITAG.

The main strength identified was that all NITAGs had developed recommendations that MoHs accepted and the majority of the recommendations were implemented. NITAG recommendations have led to the introduction of new lifesaving vaccines and the reduction of immunization inequities in the Region. Such recommendations included human papillomavirus (HPV) vaccine introduction and national strategies for COVID-19 vaccination, some of which have also been published (9, 18, 19). Two NITAGs indicated that some of their recommendations were not implemented, but the MoH did not always communicate the reasons to the NITAG. NITAG recommendations are only publicly available in three countries. In Albania and the Republic of Moldova, recommendations are published upon the MoH's decision. In Albania and Kazakhstan, interested bodies can access recommendations upon request.

3.1.7 Integration into policy processes

All NITAGs are recognized by national stakeholders, but two NITAGs indicated a lack of public recognition. Three NITAGs do not regularly consult with other NITAGs or participate in NITAG Networks (e.g., Global NITAG Network), whereas two NITAGs have interacted directly with other NITAGs.

3.2 Improvement plan development and implementation

With the project team's support, six of the evaluated NITAGs have developed improvement plans based on the provided recommendations. The improvement plans included revising the NITAGs' ToR to include important aspects of NITAG functioning, adapting the ToR to reflect current NITAG practices, or developing an SOP. In the remaining three countries, developing improvement plans were delayed due to capacity limitations; however, the project team continues working with the remaining three NITAGs to develop and implement improvement plans.

The project team supported the implementation of the NITAG improvement plans by developing specific tools and templates for NITAGs. To support NITAGs in implementing a systematic approach for evidence-based recommendation-making, guidance on an adapted EtR process for NITAGs was developed that acknowledged the human resource constraints within the Secretariats (20). In 2022 and 2023, hands-on NITAG training was conducted with the NITAGs of Armenia, Bosnia and Herzegovina, Republic of Moldova, Serbia, and Uzbekistan to apply the adapted EtR process to a specific policy question during periodic webinars with the NITAG WGs targeting each step of the process, resulting in a systematically developed vaccination recommendation.

The Regional Office has published a NITAG ToR template (21) and a WG ToR template will be published soon.

4 Discussion

The evaluations allowed NITAGs to review their composition, functioning, and quality of work outputs. Areas for further improvement were identified and reasons for existing challenges were revealed. The evaluations allowed the Regional Office and NITAG partners in the Region to gain an in-depth understanding of the NITAGs' functioning and challenges to tailor support activities to the NITAGs' needs. When we compared the current results with those from previous evaluations conducted in MICs (e.g., SIVAC evaluation), we found that the majority of challenges identified in the past (e.g., lack of a comprehensive SOP, CoI policy, standardized recommendation framework, annual work plan, use of working groups) persist and have not been completely resolved in recent years. This reveals the importance of developing improvement plans based on the evaluation findings and stringent follow-up and support of NITAGs to allow for their implementation.

The development of NITAG improvement plans, informed by evaluation findings and recommendations, along with partners' support in their implementation, has significantly contributed to strengthening the evaluated NITAGs. NITAGs enhanced their composition and functioning by developing or revising NITAG charters and ToRs to align them with best practices and WHO recommendations. After participating in EtR training sessions, NITAGs improved and standardized their recommendation-making process by integrating the EtR process into their routine practice, ensuring that their scientific advice is based on the best available evidence.

Strengthening NITAGs' capacity through evaluation and the implementation of improvement plans plays an important role in promoting equitable vaccine access. By enhancing NITAGs, we ensure that they provide robust evidence-based recommendations that lead to more informed decision-making by MoHs on introducing new vaccines, thereby contributing to equal access to life-saving vaccines in all countries. Improved NITAG capacity helps to thoroughly consider and address potential barriers to equitable access to recommended interventions, ensuring that all population groups have equal access to life-saving vaccines. Furthermore, well-functioning NITAGs increase the credibility and public trust in MoH decisions, which is essential for increasing vaccine acceptance and uptake. These efforts also contribute to achieving the European Immunization Agenda 2030's goal of increasing equitable access to new and existing vaccines for everyone (22). By ensuring that all countries have the capacity to make informed, evidence-based decisions about immunization, we move closer to achieving universal vaccine coverage and protecting public health on a global scale. NITAGs reported that conducting self-evaluations and implementing improvement plans required significant time and human resources, delaying the project's implementation. In the future, the project team should make greater efforts to motivate and incentivize NITAGs to participate in the evaluations. Sharing experiences and best practices between NITAGs regarding previous evaluations and improvement plans may be instrumental in overcoming this challenge.

A major challenge in implementing NITAG improvement plans and applying a systematic approach to the routine NITAG decision-making process was the lack of dedicated Secretariat staff. Personnel from NPHIs or NIPs, who serve as Secretariat for all evaluated NITAGs, have limited capacity because they manage Secretariat responsibilities alongside their primary job duties. WHO and NITAG partners should continue to advocate to MoHs for increased support of NITAGs, including financial support, to enable the provision of dedicated staff to serve as the Secretariat.

The Regional Office and RKI plan to conduct evaluations of the remaining seven MIC NITAGs in the Region and execute their improvement plans in 2024–2025. Based on the team's experience, it's important to have continuous follow-ups with NITAGs on the evaluation and the development and implementation of improvement plans. The experiences learned from countries that have already implemented improvement plans as well as the resources developed in the past years will make the development and implementation of future country plans easier. Additional training sessions on the adapted EtR process will be conducted and the training format will be adapted based on the results of a planned evaluation. NITAG evaluations will be repeated post-implementation to assess progress and identify areas requiring further enhancement.

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

WK-S: Conceptualization, Data curation, Formal analysis, Methodology, Visualization, Writing – review & editing, Project administration, Writing – original draft. LM: Conceptualization, Methodology, Project administration, Writing – review & editing. LJ-C: Writing – original draft, Writing – review & editing, Conceptualization. AF: Writing – review & editing. TH: Writing – review & editing. EK: Writing – review & editing. IP: Writing – review & editing. AB: Writing – review & editing. GS: Writing – review & editing. OR: Writing – review & editing. VS: Writing – review & editing. SM: Writing – review & editing. DB: Writing – review & editing. NA: Writing – review & editing. ZN: Writing – review & editing. VT: Writing – review & editing. NR: Writing – review & editing. VG: Writing – review & editing. LM-D: Writing – review & editing. BB-N: Writing – review & editing. DT: Writing – review & editing. NT: Writing – review & editing. OW: Supervision, Writing – review & editing. SD: Supervision, Writing – review & editing.

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Conflict of interest

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

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Supplementary material

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EDITED BY
Olatunde Aremu,
Birmingham City University, United Kingdom

REVIEWED BY
Yara Halasa-Rappel,
University of Massachusetts Medical School, United States
Barbara Sawicka,
University of Life Sciences of Lublin, Poland

*CORRESPONDENCE
Wei Pan
✉ weipan1977fs@163.com
Jinqing He
✉ hejinqing1987@126.com

†These authors have contributed equally to this work

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Global, regional, and national trends in pulmonary arterial hypertension burden, 1990–2021: findings from the global burden of disease study 2021

Zhenhao Liu[1†], Liumei Mo[2,3†], Wenjing Cao[2,3†], Kui Wang[4†], Hanxian Gong[2,3], Chen Li[1], Wei Pan[2,3]* and Jinqing He[5]*

[1]Department of Cardiovascular Medicine, Pingxiang People's Hospital, Jiangxi, China, [2]Department of Cardiology, Foshan Women and Children Hospital, Foshan, China, [3]Department of Geriatrics, Foshan Women and Children Hospital, Foshan, China, [4]The First Clinical Medical College, Qilu Hospital of Shandong University, Jinan, China, [5]Department of Cardiology, Ganzhou People's Hospital, Ganzhou, China

Objectives: Pulmonary arterial hypertension (PAH) is a severe and life-threatening condition. This study systematically examines the global epidemiology of PAH, focusing on trends in incidence, mortality, and disability-adjusted life years (DALYs) over the past 32 years to inform evidence-based policy and healthcare strategies.

Methods: Data from the Global Burden of Disease (GBD) 2021 study was used to analyze PAH incidence, mortality, and DALYs globally, regionally, and nationally from 1990 to 2021. Age-standardized incidence rate (ASIR), death rate (ASMR), disability-adjusted life years rate (ASDR) and estimated annual percentage change (EAPC) were assessed by age, gender, and socio-demographic index (SDI) quintiles. Hierarchical cluster analysis was performed to evaluate the temporal patterns of disease burden changes across GBD regions.

Results: Global PAH incident cases increased by 85.6%, from 23,301 in 1990 to 43,251 in 2021. ASIR increased slightly from 0.50 to 0.52 per 100,000 persons (EAPC 0.05%). From 1990 to 2021, PAH-related deaths increased from 14,842 to 22,021, though ASMR decreased (EAPC -0.57%). In 2021, PAH accounted for 642,104 DALYs, with ASDR showing a downward trend (EAPC -1.31%). Regions with low SDI exhibited the highest ASIR, while both ASMR and DALYs decreased across all SDI categories. Southern Sub-Saharan Africa had the highest incidence, while Central Asia saw the largest increases in mortality and DALYs.

Conclusion: Over the past 32 years, global ASMR and ASDR for PAH have decreased, while ASIR showed a modest increase. Persistent imbalances in treatment and outcomes remain in certain regions. Enhanced prevention and comprehensive management strategies are needed to diminish the global PAH burden and improve health equity.

KEYWORDS

pulmonary arterial hypertension, disease burden, disability-adjusted life years, incidence, death

Introduction

Pulmonary arterial hypertension (PAH) is a severe disorder characterized by elevated pulmonary artery pressure resulting from increased pulmonary vascular resistance (1). Without appropriate treatment, PAH typically progresses to right ventricular failure and ultimately leads to mortality, contributing to high morbidity and mortality rates (2). The global disease burden of PAH is rapidly evolving due to advances in medical diagnostics and treatments, accelerated ageing of the global population, and the multifaceted influence of socio-economic factors such as dietary habits and exposures to occupational and environmental toxins. Since its initial characterization, the demographic profile of PAH patients has shifted, with younger women of childbearing age now recognized as the prototypical individuals with idiopathic PAH. Changes in estrogen levels and its metabolic pathways are conncened with an increased risk of PAH in females, highlighting the hormone's modulatory role in disease susceptibility (3, 4).

Despite extensive research, the precise pathophysiologic mechanisms of PAH remain unclear (5–7). PAH encompasses a spectrum of pathophysiological phenotypes, including pulmonary artery smooth muscle cell (PASMC) hyperproliferation, pulmonary artery endothelial cell (PAEC) dysfunction, metabolic reprogramming (such as the Warburg effect), impaired angiogenesis, resistance to apoptotic resistance, chronic inflammation, and phenotypic plasticity (8, 9). The current clinical guidelines are based on these pathophysiological mechanisms and recommend combination therapies (10, 11, 35, 36). Despite notable advancements in diagnostic and therapeutic modalities in recent years, there remains a critical need for a deeper comprehension of the global disease burden of PAH to better understand its specific impact on public health. Which continues to represent a significant global health issue, necessitating ongoing research and enhanced public health strategies to mitigate its impact.

To provide further epidemiological evidence, elucidate the progress in disease health management, offer relevant message on disease focused interventions, and help different countries develop targeted strategies for prevention and control, a comprehensive and long-term analysis of PAH burden is necessary. In view of this, we conducted a comprehensive analysis using the Global Burden of Disease (GBD) 2021 dataset. Our objective was to evaluate the trends in PAH incidence, mortality, and disability-adjusted life years (DALYs) from 1990 to 2021 at global, regional, and national levels. In order to identify populations most affected by PAH and to encourage the development of appropriate prevention and treatment strategies, the studies were also stratified by age, gender, and Socio-Demographic Index (SDI).

Methods

Data acquisition and download

This study utilized data from the GBD 2021 study, which provides a wide-ranging assessment of health loss attributable to 371 diseases, injuries, and conditions, as well as 88 risk factors, across 204 countries and 811 subnational locations (12). The GBD 2021 database encompasses extensive epidemiological data, and employs advanced standardized methodologies to generate robust and comparable health estimates (12). In current study, we extracted data on the incidence, mortality, and DALYs related to PAH, as well as age, location, and sex-specific numbers and rates were calculated through the Global Health Data Exchange (GHDx) query tool.[1] Notably, The indicators included in the GBD 2021 report were defined by estimates and their 95% uncertainty intervals (UIs)., which were defined by the 25th and 75th ordered values of 1,000 draw of the posterior distribution, and all ratios were reported by every 100,000 persons according to GBD's algorithm.

The PAH-specific data, including the SDI, were obtained from online.[2] The SDI scores range from 0 to 1 and represent a country's level of social development, encompassing factors such as income per capita, educational attainment, and fertility rates (13).

Case definition

Pulmonary hypertension is classified into 5 groups (5). In the GBD 2021, PAH was classified as Group 1 pulmonary hypertension, is a vascular disease characterized by pulmonary artery remodeling, elevated pulmonary pressures, and eventual right heart dysfunction.[3] The GBD study defined PAH cases based on clinical diagnosis supported by evidence from either right heart catheterization or echocardiography. Cases identified through 10th iteration of the International Classification of Disease (ICD-10) codes 416 and I27.0 were included if diagnoses were confirmed via medical record review. To ensure specificity, ICD code I27.2 and data from ICD8, ICD9 BTL, and ICD10 tabulated formats were excluded due to their inability to reliably distinguish PAH from other pulmonary diseases. In countries where the introduction of I27.0 significantly altered PAH mortality trends, data before the implementation of I27.0 were excluded. This approach was designed to focus exclusively on PAH while excluding pulmonary hypertension groups 2–5, which have distinct pathophysiology and causes (14).

Statistics analysis

Extensive prior research has thoroughly delineated the methodologies and protocols utilized in GBD studies (15). In this research, the burden of incidence, mortality, and DALYs during the study period was evaluated by age-standardized incidence rate (ASIR), age-standardized death rate (ASMR), age-standardized DALYs rate (ASDR), and estimated annual percentage changes (EAPC) from 1990 to 2021.

Estimated Annual Percentage Change (EAPC) is a widely used metric for examining the temporal trends in disease burden over specific periods. The EAPC was calculated from the formula: $y = \alpha + \beta x + \varepsilon$, where y represents the natural logarithm of the age-standardized rate (ASR), and x denotes the calendar year. The EAPC is then calculated as $100 \times [\exp(\beta) - 1]$. The presence of a positive EAPC with its 95% confidence intervals (CIs) is an indicator that there is an upward trend in ASR, whereas a negative EAPC and its corresponding 95% CIs suggest a downward trend. Using the EAPC values, hierarchical cluster analysis was conducted to assess the temporal patterns of disease burden changes across GBD regions, thereby identifying regions with similar trends (16). Additionally, given that the variables were not normally distributed. Smoothing splines models were used to evaluate the relationship between

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[1] https://vizhub.healthdata.org/gbd-results/

[2] https://ghdx.healthdata.org/gbd-2021

[3] https://www.healthdata.org/gbd/methods-appendices-2021

the burdens of age standardized rate and EAPC in 204 countries and territories. We fitted smooth splines using the Locally Weighted Scatterplot Smoothing (LOWESS) method, which automatically determines the degree, number, and location of nodes (knots) on the basis of the data and the span parameter (17).

The association between the burden of PAH and the SDI across 21 regions and 204 countries or territories was analyzed using smoothing spline models. Expected values were derived based on the SDI and age-standardized rates across all locations. To further assess this relationship, Locally Weighted Scatterplot Smoothing (LOWESS) and Spearman correlation were applied to calculate the R indices and *p*-values, capturing the strength and significance of the correlation between age-standardized rates and SDI (18).

Additionally, subgroup analyses were performed for specific age groups to identify differential trends across different demographics. Previous studies have demonstrated that BAPC model outperforms alternative prediction models in terms of coverage and accuracy, thereby offering enhanced reliability and precision in statistical estimation. All statistical analyses were performed using R software (version 4.4.1). All hypothesis tests were two-tailed with a significance level of $p < 0.05$.

Ethical considerations

As this study utilized publicly available, de-identified data from the GBD 2021 study, ethical approval was not required. However, all analyses were conducted in accordance with relevant ethical guidelines and data usage policies.

Results

Global level

The global burden of PAH remained considerable, with the number of incident cases of PAH increased by 85.6%, from 23,301 (95% UI, 19,037-27,809) in 1990 to 43,251 (95% UI, 34,705-52,441) in 2021 (Table 1). The ASIR showed a slight increase from 0.50 per 100,000 persons in 1990 to 0.52 per 100,000 population in 2021, with an EAPC of 0.05 (95% CI: 0.03–0.07), indicating a stable trend (Table 1; Figure 1A). Globally, PAH-related deaths increased from 14,842 (95% UI: 12,370–17,485) in 1990 to 22,021 (95% UI: 18,239–25,352) in 2021 (Table 2). The

TABLE 1 Incidence of Pulmonary Arterial Hypertension between 1990 and 2021 at the global and regional level.

Location	All-age cases (95% UI)		ASIR (95% UI)		EAPC_95CI 1990−2021
	1990	2021	1990	2021	
Global	23,301 (19,037, 27,809)	43,251 (34,705, 52,441)	0.5 (0.4, 0.6)	0.52 (0.42, 0.62)	0.05 (0.03 to 0.07)
High-middle SDI	4,469 (3,599, 5,419)	7,734 (6,187, 9,586)	0.43 (0.35, 0.52)	0.46 (0.37, 0.55)	0.03 (−0.05 to 0.12)
Low SDI	2,749 (2,266, 3,274)	5,712 (4,685, 6,847)	0.78 (0.64, 0.93)	0.71 (0.58 0.85)	−0.3 (−0.36 to −0.25)
High SDI	3,677 (2,945, 4,509)	5,612 (4,500, 6,959)	0.37 (0.3, 0.45)	0.37 (0.29, 0.44)	−0.06 (−0.09 to −0.04)
Low-middle SDI	5,120 (4,183, 6,102)	9,984 (8,086, 12,020)	0.6 (0.48, 0.71)	0.59 (0.47, 0.7)	0.03 (−0.05 to 0.12)
Middle SDI	7,264 (5,922, 8,765)	14,174 (11,319, 17,383)	0.54 (0.43, 0.64)	0.53 (0.43, 0.64)	−0.17 (−0.2 to −0.13)
Andean Latin America	151 (124, 182)	347 (282, 418)	0.53 (0.43, 0.63)	0.33 (0.26, 0.4)	0.14 (0.05 to 0.22)
Australasia	77 (62, 94)	153 (123, 190)	0.35 (0.28, 0.43)	0.44 (0.36, 0.54)	0.18 (0.14 to 0.21)
Caribbean	137 (112, 166)	247 (199, 299)	0.45 (0.37, 0.55)	0.5 (0.4, 0.6)	0.27 (0.19 to 0.36)
Central Asia	233 (190, 279)	399 (320, 487)	0.41 (0.33, 0.5)	0.56 (0.45, 0.67)	0.09 (−0.01 to 0.18)
Central Europe	537 (430, 657)	794 (644, 986)	0.39 (0.31, 0.47)	0.58 (0.47, 0.69)	0.4 (0.28 to 0.53)
Central Latin America	679 (556, 818)	1,267 (1,024, 1,535)	0.56 (0.46, 0.68)	0.37 (0.3, 0.45)	−0.23 (−0.39 to −0.07)
Central Sub-Saharan Africa	302 (247, 363)	776 (636, 938)	0.81 (0.65, 0.97)	0.48 (0.39, 0.58)	0.27 (0.1 to 0.44)
East Asia	5,295 (4,275, 6,415)	9,572 (7,599, 11,898)	0.51 (0.41, 0.61)	0.47 (0.38, 0.57)	−0.12 (−0.16 to −0.07)
Eastern Europe	941 (757, 1,162)	1,198 (962,1,488)	0.37 (0.3, 0.45)	0.43 (0.35, 0.52)	0.22 (−0.07 to 0.5)
Eastern Sub-Saharan Africa	1,251 (1,036, 1,497)	2,687 (2,209, 3,243)	0.99 (0.81, 1.17)	0.41 (0.33, 0.49)	−0.07 (−0.15 to 0.01)
High-income Asia Pacific	595 (475, 733)	942 (761, 1,175)	0.31 (0.25, 0.37)	0.55 (0.44, 0.66)	0.18 (0.14 to 0.22)
High-income North America	845 (683, 1,031)	1,524 (1,209, 1,891)	0.27 (0.22, 0.33)	0.49 (0.4, 0.59)	0.35 (0.27 to 0.42)
North Africa and Middle East	1,353 (1,106, 1,612)	2,881 (2,326, 3,524)	0.56 (0.45, 0.67)	0.33 (0.27, 0.41)	−0.45 (−0.62 to −0.28)
Oceania	26 (21, 31)	65 (53, 79)	0.57 (0.46, 0.68)	0.49 (0.39, 0.59)	−0.03 (−0.12 to 0.07)
South Asia	4,573 (3,718, 5,444)	9,520 (7,651, 11,461)	0.56 (0.46, 0.67)	0.52 (0.42, 0.63)	−0.07 (−0.1 to −0.04)
Southeast Asia	1,922 (1,567, 2,311)	4,082 (3,268, 4,966)	0.55 (0.44, 0.66)	0.3 (0.24, 0.37)	0.08 (0.03 to 0.13)
Southern Latin America	162 (132, 198)	259 (208, 320)	0.34 (0.28, 0.41)	0.61 (0.49, 0.73)	0.02 (−0.02 to 0.06)
Southern Sub-Saharan Africa	317 (260,3 78)	537 (439, 649)	0.81 (0.67, 0.97)	0.83 (0.67, 0.98)	−0.08 (−0.17 to 0.01)
Tropical Latin America	602 (488, 725)	1,226 (984, 1,492)	0.5 (0.4, 0.6)	0.92 (0.75, 1.09)	−0.07 (−0.19 to 0.06)
Western Europe	2,176 (1,739, 2,656)	2,593 (2,092, 3,209)	0.46 (0.37, 0.55)	0.75 (0.61, 0.9)	−0.42 (−0.48 to −0.35)
Western Sub-Saharan Africa	1,128 (931, 1,343)	2,181 (1,786, 2,634)	0.83 (0.68, 0.99)	0.64 (0.52, 0.78)	−1.15 (−1.27 to −1.02)

ASIR, Age standardized incidence rate; EAPC, estimated annual percentage change; SDI, socio-demographic index; 95% UI, 95% uncertainty interval; 95% CI, 95% confidence interval.

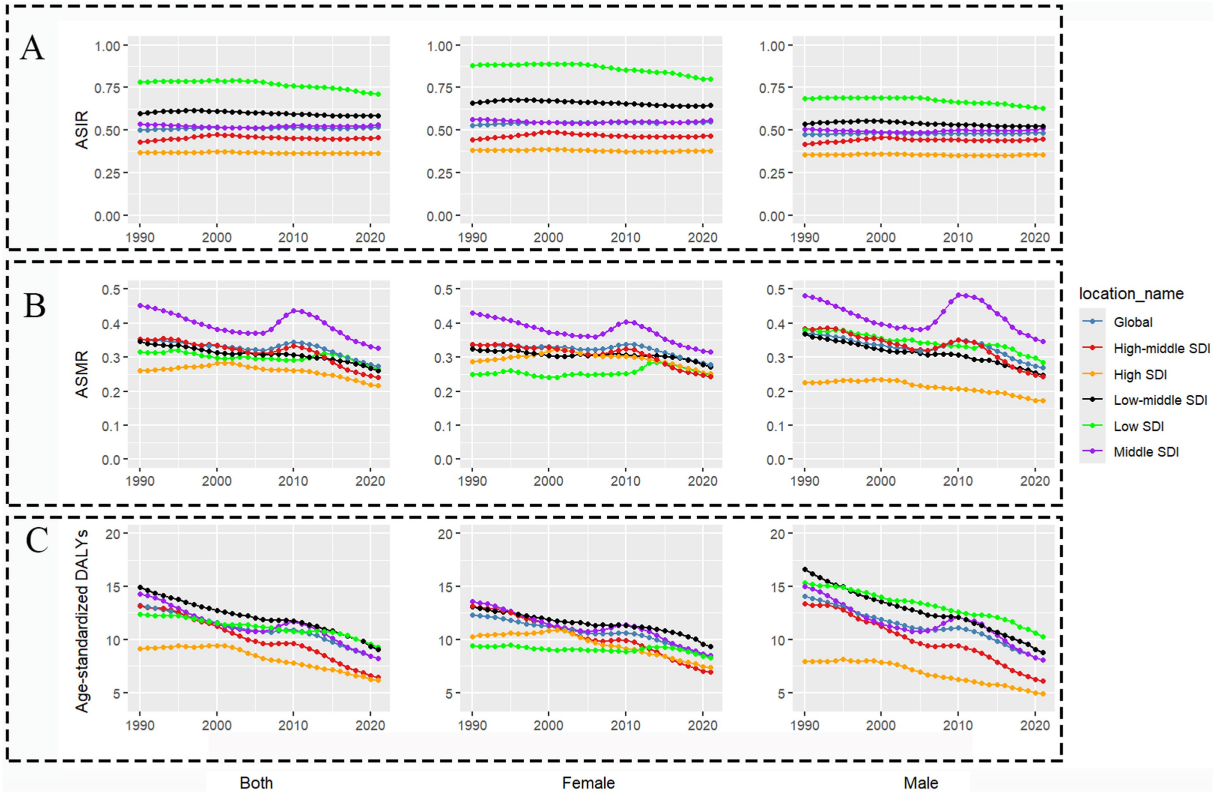

FIGURE 1
Trends in pulmonary arterial hypertension ASIR, ASMR and ASDRfrom 1990 to 2021. **(A)** ASIR. **(B)** ASMR. **(C)** ASDR. ASIR, age-standardized incidence rate; ASMR, age-standardized death rate; DALYs, disability-adjusted life years; ASDR, age-standardized DALYs rate.

age-standardized mortality rate (ASMR) was 0.27 per 100,000 persons (95% UI: 0.23–0.32), with an EAPC of −0.57 (95% CI: −0.72 to −0.42), indicating a decreasing trend in ASMR (Table 2; Figure 1B). In 2021, PAH accounted for 642,104 DALYs (95% UI: 552,273–728,993), with an ASDR of 8.24 per 100,000 persons (95% UI: 7.14–9.39), with an EAPC of −1.31 (95% CI: −1.43 to −1.19), reflecting a decrease in ASDR (Table 3; Figure 1C).

Regional level

The global burden of PAH exhibits notable variations that are closely associated with SDI levels across regions. At the SDI region level, the ASIR exhibited notable inconsistencies. From 1990 to 2021, low SDI regions had the highest ASIR decreasing from 0.78 to 0.71 per 100,000 persons, with an EAPC of −0.3% (95% CI: −0.36% to −0.25%), representing the most substantial decrease (Table 1; Figure 1A). In contrast, ASIR increased in the low-middle and high-middle SDI regions, while it decreased was observed in the remaining three regions. High SDI regions exhibited the lowest ASMR and ASDR, whereas low SDI regions recorded the highest ASDR (Table 3). Specifically, middle SDI regions reported the highest ASMR at 0.33 (95% UI: 0.22–0.39) compared to 0.22 per 100,000 persons (95% UI: 0.19–0.23) in high SDI regions (Table 2). Conversely, the ASDR was 9.3 per 100,000 persons (95% UI: 6.08–13.2) in high-middle SDI regions and markedly lower in high SDI regions at 6.16 per 100,000 persons (95% UI: 5.76–6.49) (Table 3).

The fitted curve revealed a nonlinear relationship between the SDI and both ASMR and ASDR, characterized by a left-skewed inverted U-shaped, peaking at low SDI levels. The burden of PAH exhibited a marked increase in regions with low SDI (SDI < 0.4). In contrast, regions with higher SDI experienced a gradual decrease in PAH burden as SDI improved (Figures 2B,C).

Across the 21 GBD regions, Sub-Saharan Africa bears the highest burden of PAH incidence globally, with the top four incidence rates observed within this region. Specifically, the ASIR was highest in Southern Sub-Saharan Africa, reaching 0.92 per 100,000 persons (95% UI: 0.75–1.09), followed closely by Central Sub-Saharan Africa at 0.82 per 100,000 persons (95% UI: 0.67–0.98) (Table 1; Figure 3A). However, the most pronounced decline in ASIR occurred in Western sub-Saharan Africa (EAPC −1.15, 95%CI: −1.27 to −1.02) (Table 1; Figure 3D). Central Asia and Middle East, North Africa and Middle East and East Asia exhibited significanely high ASMRs for PAH, each exceeding 0.4 per 100,000 persons (Table 2; Figure 3B). From1990 to 2021, the ASMR of PAH increased most in Central Asia (EAPC 0.3, 95% UI: 0.06 to 0.53) and decreased most in Eastern Europe (EAPC -3.78, 95% UI: −4.18 to −3.37) (Table 2; Figure 3E). Furthermore, Central Asia was the only region showing an increasing trend. Similarly, the regions with the highest ASDR were North Africa and the Middle East (14.81 per 100,000 persons; 95% UI: 10.76–17.95), Central Asia (12.91 per 100,000 persons; 95% UI: 10.61–15.60), and the Caribbean (11.73 per 100,000 persons; 95% UI: 6.48–18.74) (Table 3; Figure 3C). From 1990 to 2021, ASDR for PAH noticeably decreased across all regions, with the most significant decrease observed in Eastern Europe

TABLE 2 Morality of Pulmonary Arterial Hypertension between 1990 and 2021 at the global and regional level.

Location	Death cases (95% UI)		ASMR (95% UI)		EAPC_95CI 1990–2021
	1990	2021	1990	2021	
Global	14,842 (12,370, 17,485)	22,021 (18,239, 25,352)	0.35 (0.29, 0.42)	0.27 (0.23, 0.32)	−0.57 (−0.72 to −0.42)
High-middle SDI	3,214 (2,772, 3,908)	4,326 (3,594, 5,141)	0.35 (0.31, 0.43)	0.24 (0.2, 0.29)	−1.07 (−1.27 to −0.87)
Low SDI	1,145 (740, 1,755)	1,782 (1,147, 2,544)	0.32 (0.15, 0.51)	0.27 (0.15, 0.4)	−0.33 (−0.44 to −0.22)
High SDI	2,617 (2,379, 2,850)	4,621 (3,919, 5,054)	0.26 (0.24, 0.28)	0.22 (0.19, 0.23)	−0.56 (−0.74 to −0.38)
Low-middle SDI	3,125 (2,220, 3,904)	3,728 (2,757, 5,091)	0.35 (0.22, 0.47)	0.26 (0.18, 0.38)	−0.71 (−0.79 to −0.63)
Middle SDI	4,729 (3,774, 5,852)	7,548 (5,141, 9,026)	0.45 (0.35, 0.58)	0.33 (0.22, 0.39)	−0.63 (−0.88 to −0.37)
Andean Latin America	84 (56, 112)	91 (72, 119)	0.28 (0.21, 0.35)	0.16 (0.12, 0.2)	−1.7 (−1.97 to −1.44)
Australasia	45 (39, 57)	58 (49, 65)	0.21 (0.18, 0.26)	0.11 (0.1, 0.13)	−2.06 (−2.49 to −1.62)
Caribbean	124 (83, 169)	95 (66, 130)	0.38 (0.28, 0.49)	0.2 (0.13, 0.29)	−2.53 (−2.78 to −2.29)
Central Asia	208 (163, 240)	319 (261, 382)	0.4 (0.31, 0.47)	0.41 (0.34, 0.48)	0.3 (0.06 to 0.53)
Central Europe	355 (309, 394)	438 (398, 479)	0.26 (0.22, 0.28)	0.21 (0.19, 0.23)	−0.71 (−0.97 to −0.45)
Central Latin America	180 (157, 210)	201 (177, 230)	0.16 (0.14, 0.19)	0.08 (0.07, 0.1)	−2.54 (−2.89 to −2.19)
Central Sub-Saharan Africa	87 (55, 163)	131 (62, 237)	0.24 (0.11, 0.47)	0.19 (0.08, 0.37)	−0.67 (−0.74 to −0.61)
East Asia	4,115 (3,141, 5,526)	7,490 (4,986, 9,266)	0.59 (0.45, 0.81)	0.41 (0.28, 0.5)	−0.57 (−0.94 to −0.19)
Eastern Europe	563 (512, 651)	278 (258, 300)	0.24 (0.22, 0.27)	0.09 (0.08, 0.1)	−3.78 (−4.18 to −3.37)
Eastern Sub-Saharan Africa	366 (217, 687)	468 (219, 878)	0.27 (0.12, 0.52)	0.18 (0.07, 0.34)	−1.46 (−1.51 to −1.42)
High-income Asia Pacific	434 (410, 459)	1,049 (826, 1,201)	0.26 (0.24, 0.27)	0.23 (0.2, 0.26)	−0.46 (−0.59 to −0.33)
High-income North America	1,064 (947, 1,167)	1,880 (1,620, 2,043)	0.32 (0.28, 0.35)	0.29 (0.26, 0.31)	−0.45 (−0.61 to −0.29)
North Africa and Middle East	2,142 (1,309, 2,739)	1,896 (1,328, 2,305)	0.77 (0.56, 1)	0.44 (0.31, 0.53)	−1.36 (−1.5 to −1.21)
Oceania	12 (8, 19)	25 (17, 43)	0.28 (0.18, 0.53)	0.24 (0.16, 0.48)	−0.54 (−0.59 to −0.49)
South Asia	2,385 (1,502, 3,418)	3,549 (2,321, 5,532)	0.31 (0.17, 0.5)	0.25 (0.16, 0.42)	−0.48 (−0.58 to −0.38)
Southeast Asia	506 (340, 1,094)	741 (525, 1,850)	0.15 (0.09, 0.43)	0.12 (0.08, 0.32)	−0.7 (−0.78 to −0.62)
Southern Latin America	169 (151, 186)	150 (138, 162)	0.36 (0.32, 0.4)	0.18 (0.17, 0.2)	−2.06 (−2.25 to −1.87)
Southern Sub-Saharan Africa	43 (32, 58)	72 (53, 86)	0.12 (0.08, 0.17)	0.11 (0.08, 0.13)	−0.03 (−0.26 to 0.2)
Tropical Latin America	394 (373, 412)	779 (714, 822)	0.37 (0.35, 0.39)	0.32 (0.29, 0.34)	−0.55 (−1.08 to −0.02)
Western Europe	1,233 (1,094, 1,380)	1,788 (1,533, 1,943)	0.24 (0.21, 0.27)	0.18 (0.16, 0.19)	−0.76 (−1.26 to −0.25)
Western Sub-Saharan Africa	335 (195, 616)	523 (306, 774)	0.25 (0.09, 0.51)	0.17 (0.07, 0.28)	−1.38 (−1.5 to −1.27)

ASMR, Age standardized death rate; EAPC, estimated annual percentage change; SDI, socio-demographic index; 95% UI, 95% uncertainty interval; 95% CI, 95% confidence interval.

(EAPC -3.99, 95% CI: −4.39 to −3.58) (Table 3; Figure 3F). Hierarchical clustering analysis revealed a marked increase age-standardized rates for deaths and DALYs was observed in Central Asia, Southern Sub-Saharan Africa, Oceania (Figure 4; Supplementary Figure 1).

National level

In 2021, Zambia, Ethiopia, and Uganda exhibit the highest ASIR of PAH, with rates of 1.06, 1.00, and 1.00 per 100,000 persons, respectively (Figure 3 (A)). In 2021, countries most affected by PAH in terms of ASMR and DALYs include Mongolia, Georgia, Tajikistan, and Mauritius. Mongolia leads in both metrics, holding the top ASMR (1.59 per 100,000 persons) and DALYs rate (43.92 per 100,000 persons). Georgia ranks second in ASMR (1.00 per 100,000 persons) and third in DALYs (27.83 per 100,000 persons). Tajikistan is third in ASMR (0.81 per 100,000 persons) and fourth in DALYs (27.43 per 100,000 persons). Mauritius ranks fifth in ASMR (0.71 per 100,000 persons) and second in DALYs (28.96 per 100,000 persons).

From 1990 to 2021, Slovakia saw the largest increase in ASIR (EAPC 0.91%), while Burkina Faso experienced the greatest decrease (EAPC -1.99%). Latvia recorded the highest rise in ASMR (EAPC 5.63%). Conversely, Puerto Rico demonstrated the most significant decreases in both ASMR (EAPC -6.64%) and DALYs (EAPC -6.72%). In contrast, Mauritius showed the largest increase in DALYs (EAPC 5.5%) (Figures 3B,C).

Age and sex patterns

Between 1990 and 2021, global patterns of incidence, mortality, and DALYs have significant transformed across various age groups, genders, and SDI levels. In 1990, lower SDI regions

TABLE 3 Disability adjusted life years of Pulmonary Arterial Hypertension between 1990 and 2021 at the global and regional level.

Location	DALYs (95% UI)		Age-standardized DALY rate (95% UI)		EAPC_95CI 1990–2021
	1990	2021	1990	2021	
Global	687,419 (535,241, 813,086)	642,104 (552,273, 728,993)	13.21 (10.78, 15.36)	8.24 (7.14, 9.39)	−1.31 (−1.43 to −1.19)
High-middle SDI	127,638 (106,711, 154,438)	99,448 (85,757, 117,639)	13.14 (10.91, 16.04)	6.48 (5.61, 7.87)	−2.2 (−2.36 to −2.05)
Low SDI	71,125 (48,628, 111,614)	95,342 (67,471, 133,050)	12.42 (7.78, 19.19)	9.3 (6.08, 13.2)	−0.78 (−0.85 to −0.71)
High SDI	81,792 (77,185, 88,110)	93,182 (84,873, 99,192)	9.16 (8.71, 9.9)	6.16 (5.76, 6.49)	−1.39 (−1.57 to −1.22)
Low-middle SDI	195,281 (117,185, 245,591)	156,400 (122,426, 194,166)	14.92 (10.74, 18.41)	9.07 (7.05, 11.6)	−1.33 (−1.43 to −1.24)
Middle SDI	210,946 (172,857, 258,349)	197,171 (148,781, 232,321)	14.29 (11.66, 17.69)	8.23 (6.26, 9.7)	−1.4 (−1.61 to −1.18)
Andean Latin America	5,049 (2,977, 7,422)	3,544 (2,795, 4,490)	11.92 (7.83, 16.43)	5.73 (4.52, 7.28)	−2.11 (−2.35 to −1.86)
Australasia	1,514 (1,337, 1,856)	1,434 (1,305, 1,563)	7.37 (6.49, 9.06)	3.67 (3.38, 3.99)	−2.33 (−2.8 to −1.86)
Caribbean	7,398 (4,170, 11,145)	5,071 (2,988, 7,877)	19.72 (11.78, 28.82)	11.73 (6.48, 18.74)	−2.01 (−2.19 to −1.83)
Central Asia	9,071 (7,195, 10,646)	11,619 (9,514, 14,202)	14.14 (11.27, 16.45)	12.91 (10.61, 15.6)	−0.33 (−0.55 to −0.11)
Central Europe	11,026 (9,784, 12,154)	10,424 (9,512, 11,459)	8.15 (7.25, 8.95)	6.05 (5.5, 6.67)	−1.01 (−1.25 to −0.78)
Central Latin America	9,966 (8,844, 11,855)	7,246 (6,391, 8,407)	6.12 (5.4, 7.16)	3 (2.63, 3.51)	−2.67 (−2.99 to −2.35)
Central Sub-Saharan Africa	5,367 (3,258, 10,525)	6,524 (3,586, 11,025)	9.09 (5.65, 17.13)	6.16 (2.94, 11.09)	−1.14 (−1.24 to −1.05)
East Asia	151,596 (117,394, 205,773)	154,740 (102,939, 190,399)	15.78 (12.34, 21.21)	8.84 (5.99, 11.01)	−1.32 (−1.68 to −0.97)
Eastern Europe	20,628 (18,781, 23,860)	8,357 (7,760, 8,996)	9.23 (8.47, 10.49)	3.32 (3.1, 3.56)	−3.99 (−4.39 to −3.58)
Eastern Sub-Saharan Africa	23,148 (14,694, 45,765)	26,605 (14,265, 49,615)	11.11 (6.18, 21.17)	6.85 (3.3, 12.62)	−1.62 (−1.67 to −1.58)
High-income Asia Pacific	18,474 (17,689,1 9,467)	19,988 (17,442, 21,997)	12.03 (11.43, 12.79)	8.31 (7.74, 8.87)	−1.5 (−1.7 to −1.29)
High-income North America	30,206 (27,736, 32,715)	38,373 (35,060, 40,844)	10.13 (9.39, 10.99)	7.71 (7.17, 8.18)	−1.14 (−1.32 to −0.95)
North Africa and Middle East	145,728 (74,879, 204,070)	80,753 (58,086, 98,810)	35.84 (21.25, 46.14)	14.81 (10.76, 17.96)	−2.43 (−2.57 to −2.29)
Oceania	704 (448, 1,090)	1,455 (988,2,411)	10.85 (7.02, 17.55)	10.14 (6.9, 17.1)	−0.22 (−0.26 to −0.17)
South Asia	136,086 (77,808, 184,381)	136,563 (97,809, 189,353)	12.24 (7.79, 17.12)	8.54 (6.02, 12.46)	−0.95 (−1.03 to −0.87)
Southeast Asia	27,786 (18,242, 51,943)	31,112 (22,913, 58,708)	6.31 (4.28, 12.72)	4.65 (3.39, 9.25)	−0.92 (−1 to −0.84)
Southern Latin America	7,983 (7,181, 8,803)	4,739 (4,439, 5,092)	16.28 (14.65, 17.95)	6.55 (6.13, 7.07)	−2.87 (−3.01 to −2.72)
Southern Sub-Saharan Africa	2,188 (1,667, 2,875)	3,212 (2,359, 3,891)	4.7 (3.65, 6.37)	4.33 (3.21, 5.2)	−0.02 (−0.26 to 0.21)
Tropical Latin America	19,065 (17,856, 20,357)	24,235 (23,002, 2 5,403)	14.18 (13.35, 14.98)	10.22 (9.65, 10.78)	−1.12 (−1.66 to −0.59)
Western Europe	34,599 (31,743, 38,281)	34,043 (31,024, 36,440)	8.27 (7.69,9.18)	5.05 (4.75, 5.32)	−1.55 (−1.99 to −1.11)
Western Sub-Saharan Africa	19,834 (13,553, 38,853)	32,071 (22,084, 47,222)	9.34 (5.22, 16.94)	6.5 (3.77, 9.47)	−1.14 (−1.27 to −1.01)

ASDR, Age standardized disability adjusted life years rate; EAPC, estimated annual percentage change; SDI, socio-demographic index; 95% UI = 95% uncertainty interval; 95% CI, 95% confidence interval.

experienced high incidence rates, particularly among children under five (Figure 5A). By 2021, incidence rates in younger populations markedly decreased, reflecting improvements in healthcare infrastructure, vaccination programs, and disease prevention initiatives. Conversely, incidence has surged among older adults, especially those aged 50 and above, in high and high-middle SDI regions (Figure 5B).

In 1990, mortality rates were predominantly high among younger populations in low SDI regions. By 2021, mortality rates for children under five has significantly decreased. In contrast, mortality rates among older adults, particularly those aged 65 and above, have increased in high SDI regions. (Supplementary Figures 2A,B). In 1990, DALYs were disproportionately high among younger populations in low SDI regions, where infectious diseases and child mortality were predominant health issues. By 2021, DALYs in younger age groups had substantially decreased, particularly in lower SDI regions. However, the distribution of DALYs has shifted toward older populations in high and high-middle SDI regions (Supplementary Figures 3A,B).

The influential factors for EAPC

We analyzed the correlation coefficients between the EAPC and ASR in 1990 (Figures 6A–C), as well as the SDI in 2021 (Figures 6D–F). Results show a positive correlation between EAPC and both ASMR ($R = 0.22$, $p < 0.001$) (Figure 6C) and ASDR ($R = 0.23$, $p < 0.001$) in 1990 (Figure 6B). Additionally, there was a slight positive correlation between EAPCs and SDI (ASIR: $R = 0.3$, $p < 0.001$) (Figure 6D). Furthermore, countries with a higher SDI showed an increasing trend in 2021, reflecting patterns previously observed across diverse SDI regions.

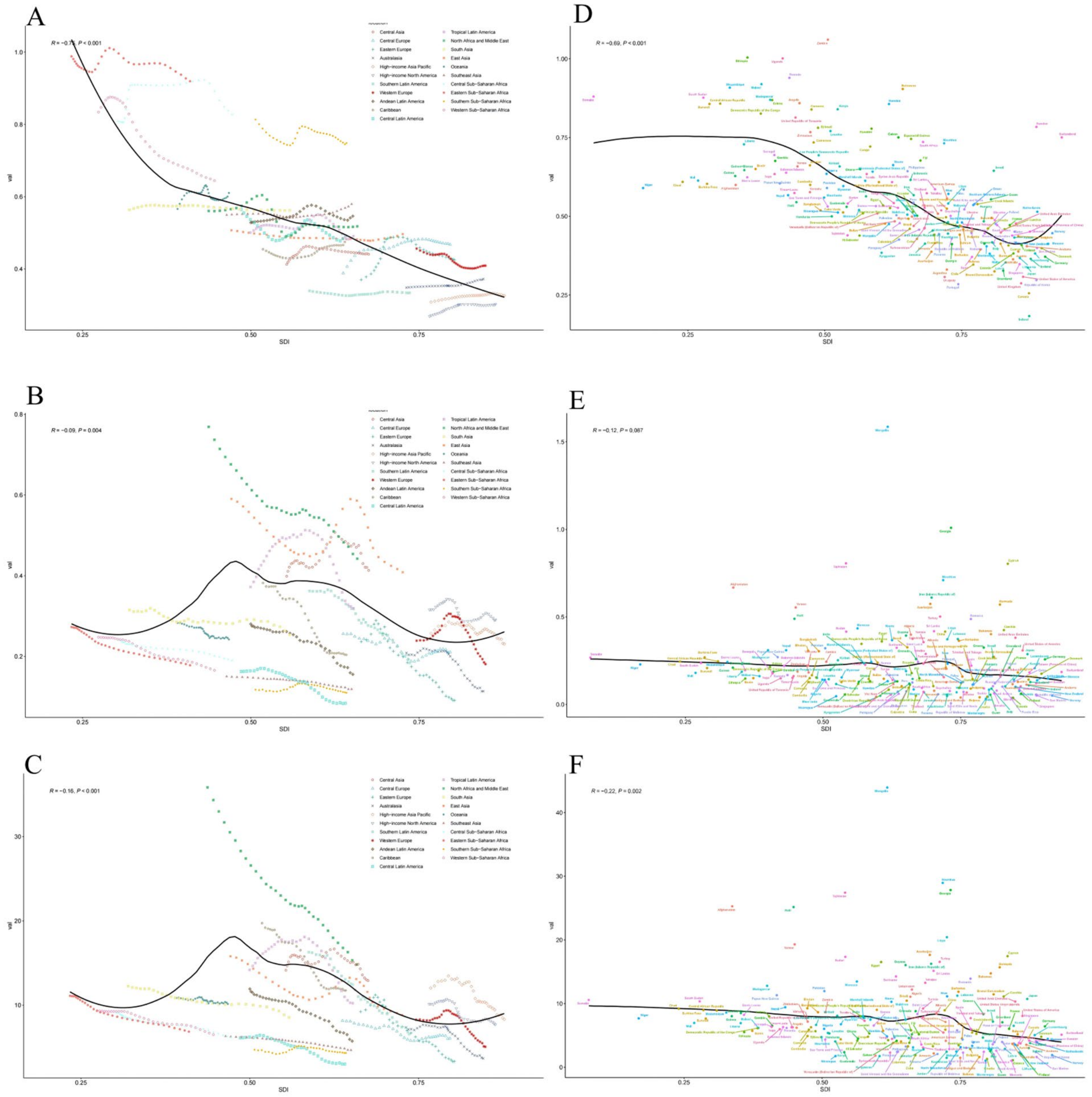

FIGURE 2
Age standardized rates of PAH among regions and nations based on SDI in 2021. **(A)** ASIR in 21 regions. **(B)** ASMR in 21 regions. **(C)** ASDRin 21 regions. **(D)** T ASIR in 204 countries. **(E)** ASMR in 204 countries. **(F)** ASDR in 204 countries. SDI, Social-Demographic Index; ASIR, age-standardized incidence rate; DALYs, disability-adjusted life years; ASDR, age-standardized DALYs rate; ASMR, age-standardized death rate.

Discussion

Previous studies have attempted to quantify the disease burden associated with PAH; however, most investigations have been limited to specific regions or countries (19, 20). Only a few studies have explored this issue on a global scale. Additionally, cohort studies confined to local populations may suffer from methodological limitations that compromise their precision. In contrast, the GBD 2021 study utilized diverse data sources, including household surveys, vital statistics, and various databases, and encompassed a wider range of countries. This approach provides more comprehensive and accurate estimates of the disease burden. Our findings offer valuable insights into the evolving burden of PAH over the past 32 years across regions and countries with different income levels. From 1990 to 2021, the burden of PAH has increased in several regions and nations globally.

In terms of age and sex distribution, this research reveals that from 1990 to 2021, there was an increasing trend in death and DALY due to PAH globally. The disease burden was more pronounced among children under five, as well as the middle-aged and older adult population. High SDI regions, having made substantial progress in reducing infectious diseases and child mortality, now face an increasing burden of chronic diseases among older adults. In contrast,

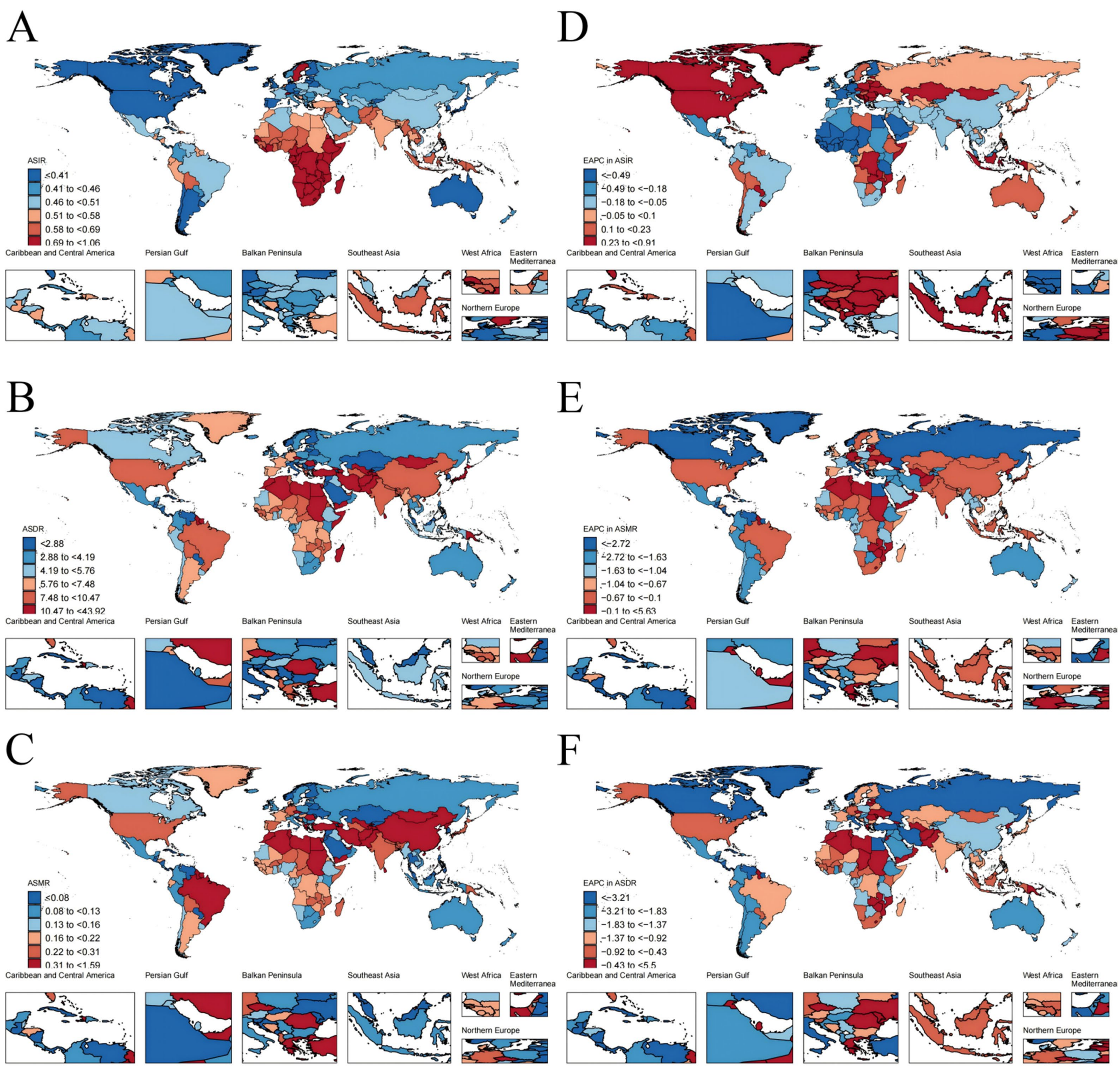

FIGURE 3
The global disease burden of Pulmonary Arterial Hypertension in 204 countries. **(A)** ASIR. **(B)** ASMR. **(C)** ASDR. **(D)** EAPC in ASIR. **(E)** EAPC in ASMR. **(F)** EAPC in ASDR. ASIR, age-standardized incidence rate; ASMR, age-standardized death rate; DALYs, disability-adjusted life years; ASDR, age-standardized DALYs rate; EAPC, estimated annual percentage change.

lower SDI regions, despite improvements in child health, continue to grapple with managing both infectious diseases and the rising incidence of chronic conditions. In summary, significant progress has been achieved in reducing incidence, mortality, and DALYs among younger populations in global, particularly children under five. However, in high SDI regions, the global health burden has increasingly shifted toward older adults. This increase is primarily attributed to the increased prevalence of non-communicable diseases such as cardiovascular disease, cancer, and diabetes, which are more common in aging populations (21, 22). This shift underscores the growing impact of chronic, age-related diseases and highlights the need for healthcare systems to adapt to the evolving demands of aging populations.

PAH has a higher prevalence in women, who also demonstrate improved prognosis, potentially attributable to oestrogen-mediated mechanisms (23, 24). Despite the overall increasing global burden of PAH over the past 32 years, mortality rates, particularly among women, have risen. Conversely, the ASMR and ASDR have shown a decreasing trend. Where chronic, non-communicable diseases significantly contribute to both morbidity and mortality. Additionally, women tend to experience higher DALYs in older age groups due to longer life expectancy and greater exposure to chronic illnesses (25). In conclusion, the gender disparity in the disease burden associated with PAH remains unclear, and the underlying mechanisms have yet to be fully elucidated (26). Therefore, comprehensive investigations are urgently needed to clarify these differences.

In terms of inter-country inequality, the disease distribution map and cross-regional comparisons indicate that the burden of PAH is closely associated with the SDI (27). This pattern likely

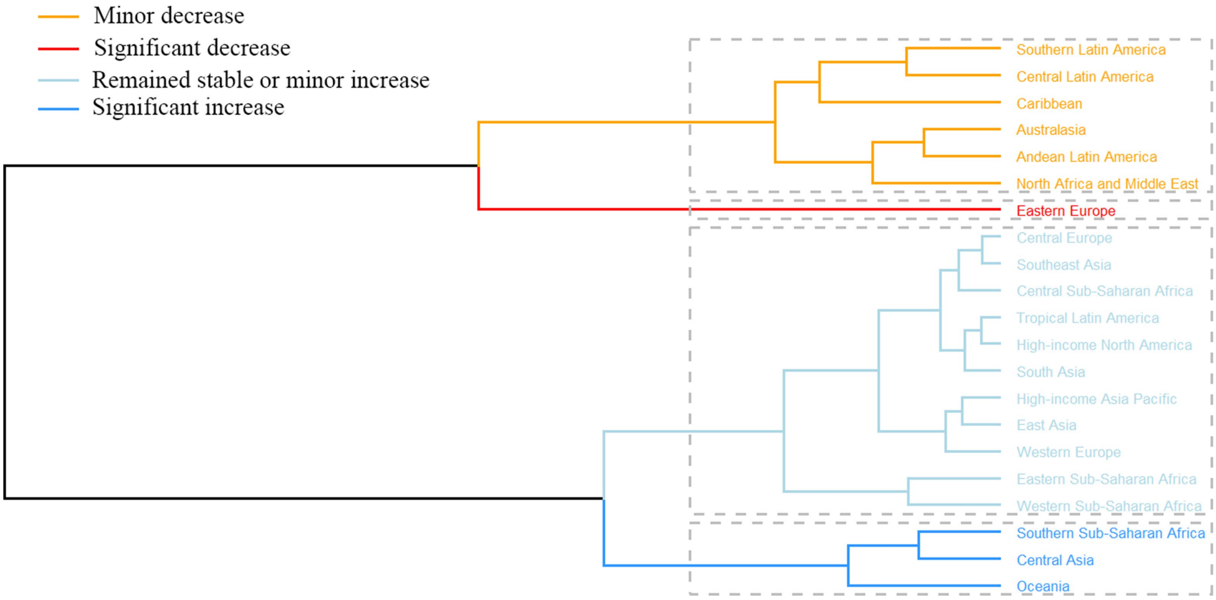

FIGURE 4
Results from clustering EAPC values for age-standardized death and DALY rates associated with PAH between 1990 and 2021. EAPC, estimated annual percentage change; PAH, pulmonary arterial hypertension; DALYs, disability-adjusted life years.

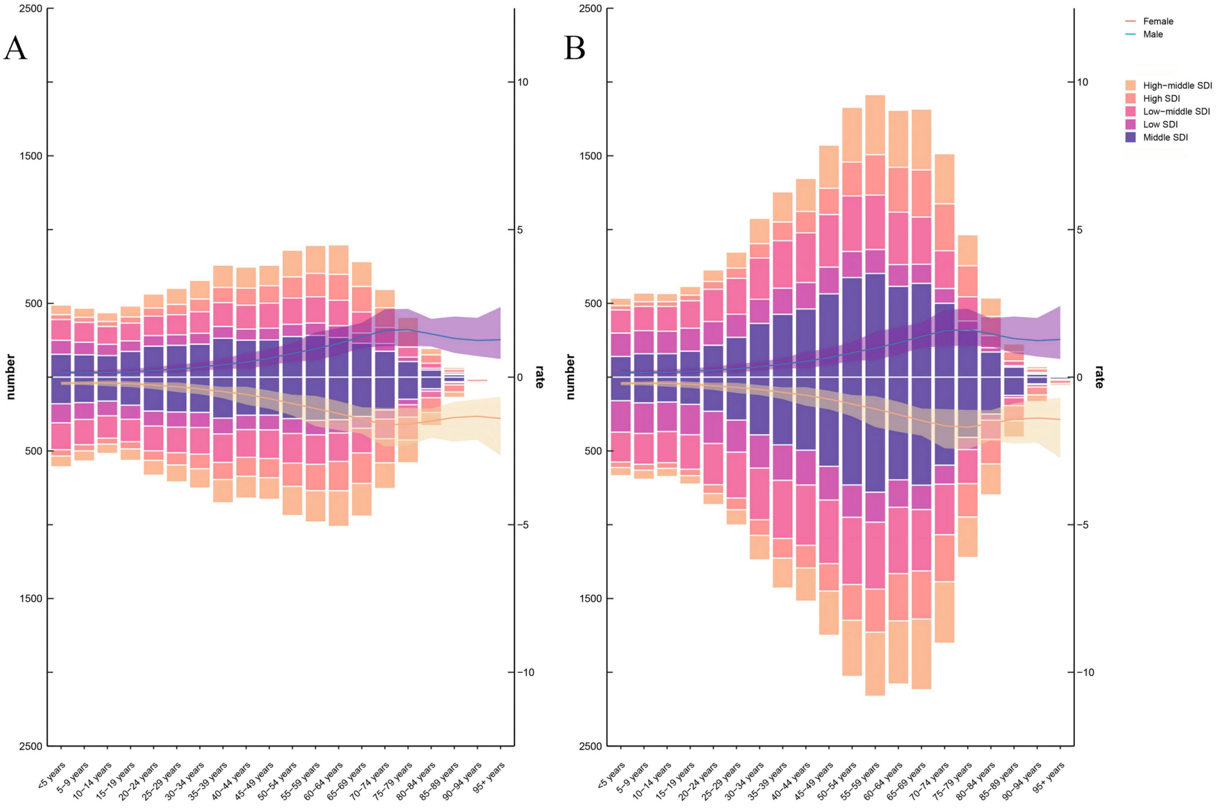

FIGURE 5
The age-specific numbers and ASIRs of PAH by SDI regions in 1990 and 2021. **(A)** ASIR in 1990. **(B)** ASIR in 2021.ASIR, age-standardized incidence rate; PAH, pulmonary arterial hypertension; SDI, Social-Demographic Index.

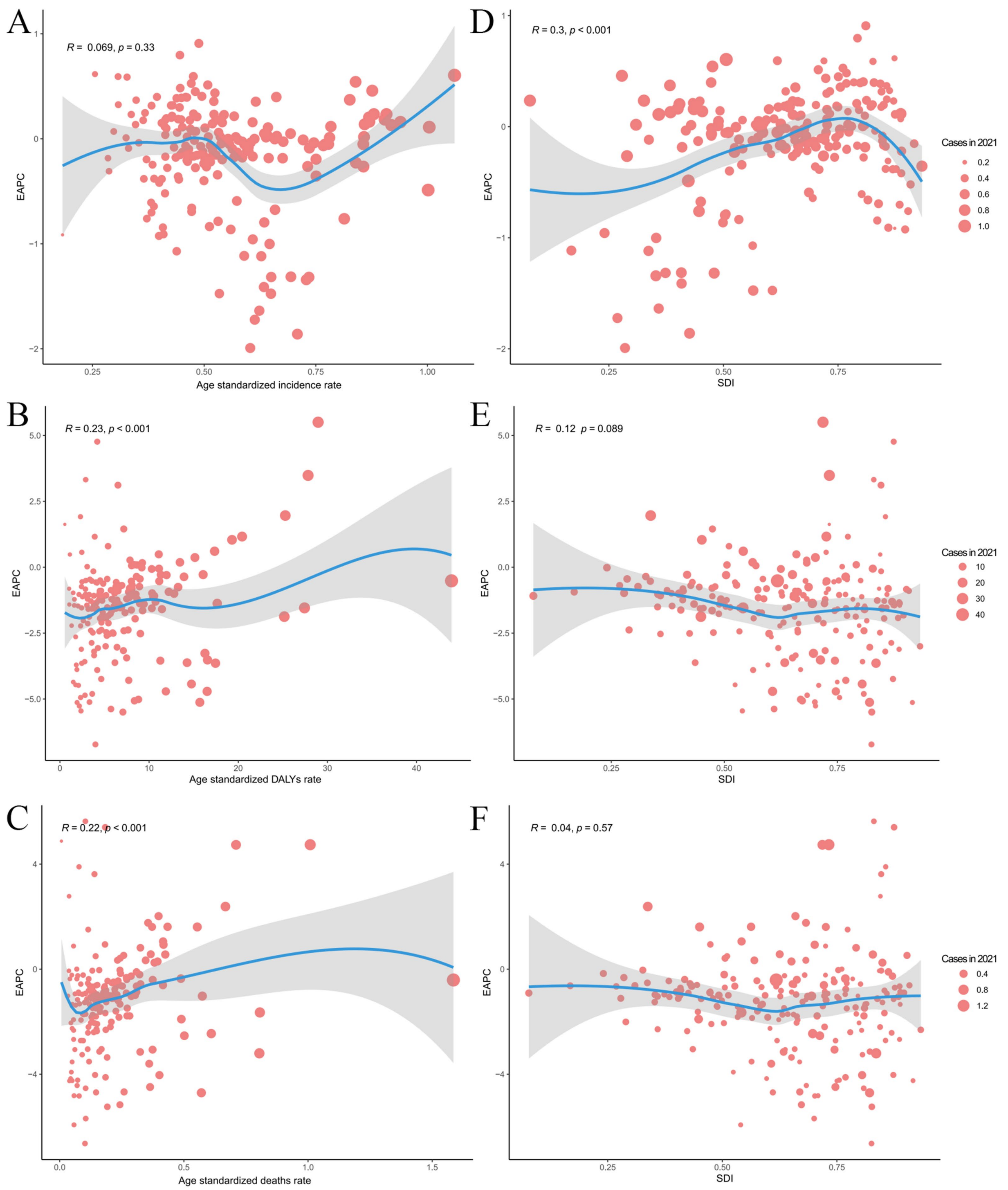

FIGURE 6
Correlation between EAPC and myocarditis ASIR, ASMR and Age-standardized disability-adjusted life-years in 1990 and SDI in 2021. **(A)** EAPC in ASIR in 1990. **(B)** EAPC in ASMR in 1990. **(C)** EAPC in ASDR in 1990. **(D)** EAPC in SDI in 2021. **(E)** EAPC in SDI in 2021. **(F)** EAPC in SDI in 2021. EAPC, estimated annual percentage change; ASIR, age-standardized incidence rate; ASMR, age-standardized death rate; SDI, Social-Demographic Index.

reflects a multifaceted interaction among variables, including an advanced diagnostic capabilities resulting in higher identification rates in developed countries, increased life expectancy broadens the population at risk in developed countries, and greater exposure to risks tied to accelerated urbanization and lifestyle shifts (28). A negative correlation was identified between PAH incidence and SDI regions. Regions with low SDI exhibited the highest ASIR, ASMR, while High SDI regions exhibiting the lowest ASIR, ASMR and ASDR. High SDI regions likely gained advantages from established healthcare infrastructure, successful public health efforts, and increased recognition of PAH risks and symptoms (29). For example, the high incidence rates in Sub-Saharan Africa and parts of Asia are probably due to a combination of genetic predisposition, HIV infection, congenital heart disease, and limitations within healthcare system (30). Central Asia experiences high mortality rates with an increasing ASM, primarily among patients with connective tissue diseases (particularly systemic sclerosis, SSc), portal hypertension, exposed to disease-causing drugs or toxins, and infected with *Schistosoma mansoni* (31, 32). Despite improvements in high SDI regions, the persistent disparity in mortality rates between low- and high-SDI areas highlights ongoing health inequities that must be addressed through targeted public health strategies and resource allocation. This underscores the intricate relationship between socio-economic factors and PAH outcomes, highlighting the impact of socio-demographic disparities on disease burden. Notably, across all SDI regions, there is an overall decline in both ASMR and ASDR.

These findings highlight the complex interplay of demographic, gender, and socioeconomic factors in shaping the global epidemiology of PAH, necessitating a multifaceted approach to disease prevention and management. However, the findings of this study ought to be interpreted with due consideration of the limitations of the study itself. Firstly, the validity of the estimates might be affected by the consistency and accessibility of data sources across diverse nations (33). In certain economically developing nations, the scarcity of accurate data on the epidemiological characteristics of PHA, combined with the insufficient cases, may result in a lower estimation of the actual global burden. Secondly, the GBD methodology is based on a number of presuppositions and computational models, which may result in a degree of inaccuracy within the produced estimates (34). Although the GBD database utilizes robust statistical techniques to mitigate these uncertainties, the findings are deemed to be the most accurate approximations currently available based on the existing evidence.

Conclusion

In conclusion, the global burden of PAH persists as a significant public health concern, exhibiting notable disparities across regions, nations, and SDI categories. These findings highlight the imperative for the development of targeted preventative and therapeutic strategies that are tailored to the specific requirements of diverse, populations. Strengthening healthcare infrastructure, encouraging healthy behaviors, and addressing socioeconomic inequalities are addressed as a means of mitigating the global impact of PAH. Future research should prioritize optimize the identification and implementation of the most effective preventative and management for PAH, with a particular focus on regions with a high prevalence of the disease. Similarly, efforts should focus on unraveling the intricate relationships between genetic predispositions, Exposures to the environment, and Lifestyle related factors that contribute to the pathogenesis of PAH.

Data availability statement

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

Ethics statement

The institutional review board of the Ganzhou People's Hospital in Jiangxi Province, China, determined that the study did not need approval because it used publicly available data.

Author contributions

ZL: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Software, Visualization, Writing – original draft, Writing – review & editing. LM: Conceptualization, Formal analysis, Investigation, Methodology, Supervision, Writing – review & editing. WC: Conceptualization, Methodology, Validation, Writing – review & editing. KW: Data curation, Software, Validation, Writing – review & editing. HG: Data curation, Formal analysis, Writing – review & editing. CL: Data curation, Formal analysis, Writing – review & editing. WP: Funding acquisition, Resources, Supervision, Writing – review & editing. JH: Project administration, Supervision, Writing – review & editing.

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Conflict of interest

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

Generative AI statement

The author(s) declare that no Gen AI was used in the creation of this manuscript.

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Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpubh.2025.1516365/full#supplementary-material

SUPPLEMENTARY FIGURE 1
Results from clustering EAPC values for age-standardized death and DALY rates associated with PAH between 1990 and 2021. EAPC, estimated annual percentage change; DALYs, disability-adjusted life years; PAH, pulmonary arterial hypertension.

SUPPLEMENTARY FIGURE 2
The age-specific numbers and ASMRs of PAH by SDI regions in 1990 and 2021. **(A)** ASMR in 1990. **(B)** ASMR in 2021. ASMR, age-standardized death rate; PAH, pulmonary arterial hypertension; SDI, Social-Demographic Index.

SUPPLEMENTARY FIGURE 3
The age-specific numbers and ASDRs of PAH by SDI regions in 1990 and 2021. **(A)** ASDR in 1990. **(B)** ASDR in 2021. DALYs, disability-adjusted life years; ASDR, age-standardized DALYs rate; PAH, pulmonary arterial hypertension; SDI, Social-Demographic Index.

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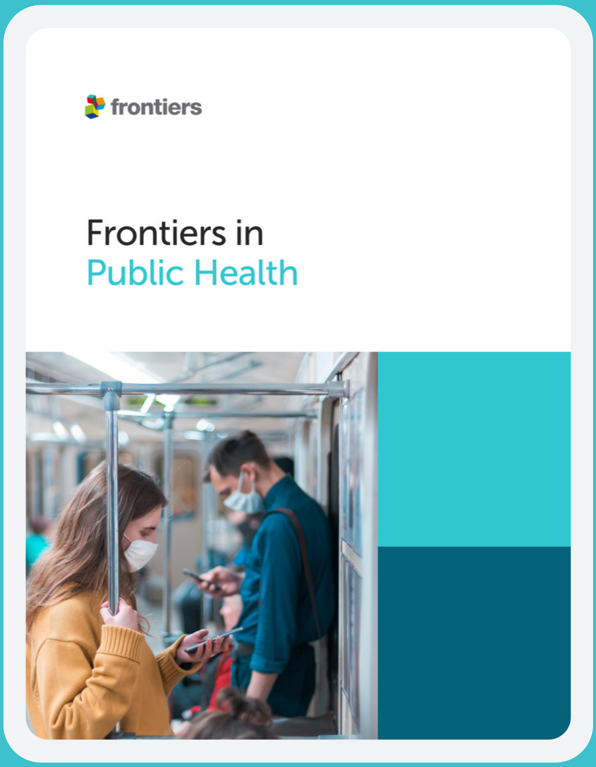
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