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COMMENTARY

Psychosocial stress and risk assessment during the COVID-19 pandemic: Some preliminary thoughts

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Abstract

Human life and activities are associated with risk, as risk is inherent in all forms of human existence and action. Risk is regarded as a phenomenological variable considering how people perceive it, especially during a crisis they experience. This paper discusses the characteristics of human behavior in a crisis, with an emphasis on hazard perception and risk assessment, for the purpose of understanding people's decisions and adaptation before, during, and after a crisis. The discussion focuses on the ongoing COVID-19 global pandemic.

Keywords: Psychosocial stress; Risk assessment; Health crisis; COVID-19; Serbia

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1. Introduction

Terms such as stress, crisis, and risk are nowadays frequently used in the scientific literature as well as in professional communication, especially now during the global coronavirus disease 2019 (COVID-19) pandemic. These terms best describe human behavior in emergencies and extreme situations. When an individual is faced with sudden, unknown, and dangerous circumstances, the first thing to emerge is a state of shock and mental and emotional inhibition, manifesting as narrowed consciousness, and sometimes even a state of catalepsy, which profoundly affects the biological sense of survival of the afflicted individual (Čabarkapa, 2016). Such responses are commonly associated with wars, severe medical conditions or injuries, earthquakes, or other "no-escape" situations due to natural or technological disasters (Seneviratne, Baldry and Pathirage, 2010).

In the majority of other stressful situations, both privately and professionally, especially in the case of psychosocial stressors, which manifest as complicated and challenging situations, a certain level of cognitive ability is retained in the earliest stage of shock and disbelief, which is why a person is able to consciously and unconsciously question what is happening and how it affects them (Biggs, Brough and Drummond, 2017). This provides an opportunity to use some of the active strategies to overcome stress, which people most often do instead of succumbing to their imminent fate of surrender and death.

The crisis caused by the uncontrolled spreading of the coronavirus has been regarded as a form of biological war against an unknown enemy, which resulted in an

extensive variety of measures undertaken to combat it. The perception of such danger caused mass worldwide stress, fear, and anxiety, which suggests that there is also a fear pandemic to consider (Vujčić, Safiye, Milikić, *et al.*, 2021). The crisis has affected all social strata, and no one has been spared from the effects.

The subject matter of this paper involves description and determination of individual behavior and risk perception when facing danger and crises. The aim of the paper is to review the specific nature and complexity of risk assessment during a crisis accompanied by stress, particularly in the case of current COVID-19 global pandemic, with an empirical focus on Serbia.

2. Psychosocial stress and risk assessment

It is a well-known fact in psychology that there is a significant difference between the actual objective state and the subjective perception of objects, events, or situations, especially when there is insufficient information or when consciousness is in an altered state, for instance under extreme stress (Čabarkapa, 2016). Under the influence of thus altered perception and a series of mental, emotional, and motivational factors affecting information processing, a wide discrepancy may arise between the objectively present risk and the subjectively perceived and assessed risk (Seneviratne, Baldry and Pathirage, 2010). This paper will not discuss all the theories, factors, and types of risk associated with different areas of people's everyday life and activity, but will refer to risk in terms of negative effects on the safety, health, and well-being of individuals, groups, or society in general within the context of stress and crisis. Hazard perception and risk assessment are an important cognitive variable for the understanding of people's decisions and their adaptation before, during, and after hazardous events (Weber, 2001; White, 1974). The previous research regarding this topic indicates that there are considerable differences in risk assessments performed by technical experts, the media and the general public, and individuals of different age, gender, and culture (Seneviratne, Baldry and Pathirage, 2010).

Important psychological determinants of perceived risk include fear, exposure, and familiarity of risks (Božović, Mihajlović and Živković, 2019). Fear refers to the possibility of suffering, which is a clear emotional and cognitive indicator of what people intuitively think of risk. Exposure refers to the actual exposure of people to hazards. Familiarity pertains to the previous experience with and knowledge about a risk. Experience gained during a disaster event significantly alters personal hazard perception as well as individual opinions and behaviors in terms of preparedness for facing danger. Memory of hazardous events and disasters usually leads to increased risk perception (Božović, Mihajlović and Živković, 2019).

Of the three said factors, fear impacts human risk perception the most. When people face danger situations, their anxiety or fear of exposure to risk impacts their behavior (Mun, Moon, Kim, *et al.*, 2021). In addition to fear, individual risk perception and assessment also depend on the *type of risk* (voluntary versus involuntary), *individual characteristics* (specific personal traits, age, level of education, income group, etc.), the *nature of consequences* (immediate versus delayed), and the *ability to control risk*. People are prone to assessing risk in a multifaceted but subjective way, which is why it is important to communicate about risk – to exchange risk information interactively among risk assessors, persons in charge, the media, stakeholders, and the broader public (Brown, 2014).

Risk perception is important in determining health protective behavior during the crisis caused by the COVID-19 pandemic (Mun, Moon, Kim, *et al.*, 2021). Public perception and social construction of risks and threats are important for analyzing, assessing, and responding to crisis situations (Borodzicz, 2005; Beck, 1992; Mun, Moon, Kim, *et al.*, 2021). Strategies and methods are being developed for the design of national or regional risk maps, with clear indications of the effects, including not only safety and health effects but also economic and social costs. Such assessments and scenarios should be instituted beforehand and used proactively to draw an adequate response from the community and hold proper political debates on risk acceptance (Borodzicz, 2005).

To explain why different people assess different risks differently, scientists proposed three paradigms: *Axiomatic*, *sociocultural*, and *psychometric* (Weber, 2001; White, 1974). Studies dealing with the axiomatic paradigm focused on the general biological and psychological principles and on the way people subjectively transform the objective information about the risk. The sociocultural paradigm is prevalently focused on culture, rather than on individual psychology, as the explanation of differences in risk decision-making. Anthropologists and sociologists claim that risk perception is rooted in cultural and social factors and that culture is essential to explaining the differences in risk perception. Studies examining the psychometric paradigm have shown that people's emotional reactions to risk events or behaviors affect their assessment of the degree of risk, as the assessment often goes beyond the objective consequences, claiming that both experts and laypersons fully perpetuate false representations of several aspects of danger (Weber, 2001; White, 1974). Researchers identified the most common systemic biases that could

justify the misconceptions concerning risk. Such biases resulted from a set of general inherent rules observed by individuals in everyday situations. These rules are technically called heuristics, and their purpose is to make complex mental tasks as simple as possible. Systemic biases that could justify the misconceptions about risk include overconfidence, desire for safety, and the conviction that something *will never happen to me/us* (Witte, Meyer, and Martell, 2001). A common misconception is the idea that, when an unfortunate event or accident occurs, it is less likely to occur for a long time afterward. People are prone to assessing risk in a multifaceted but subjective way, which is why it is important to communicate about risk – to exchange risk information interactively among risk assessors, persons in charge, the media, stakeholders, and the broader public (Witte, Meyer, and Martell, 2001).

3. The specificity of the global health crisis caused by the current covid-19 pandemic

The appearance of the coronavirus strain (SARS-CoV-2) that causes the COVID-19, which led to a pandemic by affecting almost every world country, has resulted in collective stress and a unique crisis in Serbia and the rest of the world, changing the routine of personal and family life and suspending many types of work, trade, and communication (Vujčić, Safiye, Milikić, *et al.*, 2021).

When the pandemic was officially declared, it appears that many countries were underprepared for a prompt and decisive response to the psychosocial stress and the crisis. The risk of an epidemic had initially been underestimated in most countries, but when the WHO declared a pandemic, a panic-driven race ensued to find the necessary assets, resources, and methods to control the emergent disease. Since this is a case of a biological hazard that also poses a health risk for the entire human population – not just the directly afflicted countries, decision-making regarding control measures has been entrusted to health experts – epidemiologists, virologists, infectologists, and other medical specialists. Serbia, as well as most other countries, arrived at a political and general social consensus that it is best to allow the experts to make decisions regarding risk assessment and to recommend measures for dealing with the COVID-19 crisis. Although government measures were essential for containing the spread of the COVID-19, the disrupting of a normal life during the state of emergency has proven to be a serious threat to the mental health and well-being of the general population, students and especially health care workers, as shown in the previous studies (Vujčić, Safiye, Milikić, *et al.*, 2021; Safiye and Vukčević, 2020; Safiye, Vukčević and Čabarkapa, 2021). On March 31, 2020, "Telekom," the largest mobile service provider in Serbia, sent the following text message to all users: "The situation is dramatic. We are approaching the scenarios seen in Italy and Spain. Please, stay home," which caused additional fear and increased the perception of risk among citizens (Vujčić, Safiye, Milikić, *et al.*, 2021).

However, risk assessment is not as simple as one might initially assume, especially when one considers all aspects of risk – types, levels, and consequences. Experts are expected to formulate clear, unequivocal, and depoliticized conclusions, on the basis of which public policies are then formed. When a situation is complex and it threatens to have serious consequences for the individuals and the society, the question arises if epidemiologists are currently the only ones who are competent to develop public policies and what is currently understood as professional expertise, which is called on by the political decision-makers. Does professional expertise also involve economists, psychologists, sociologists, statisticians, and other experts, who could form a multidisciplinary team and provide answers on crisis communication strategies, change management, and assessments of health, psychological, and economic losses in the different ways public policies are carried out?

Complex and dynamic states necessitate a systemic, active, contextual, and multidisciplinary approach instead of the bureaucratic implementation of previous practices. Occasionally, previous good practice can prove successful in dealing with certain issues, for instance in containing infectious diseases in China and in Serbia, but it became clear that in every crisis, the available information and resources need to be adjusted to the local context, which should be accompanied by a vigorous approach and the preparedness to take certain risks. Therefore, in addition to public policy makers, crisis management teams and expert teams should also include top experts from different fields, who could ensure that the dynamic and complex processes are dealt with vigorously and according to the best available data. Comparative studies of the effects of different strategies in different countries, focusing on the advantages and disadvantages of different models currently in use (e.g., what can be learned from China, Germany, Sweden, Italy, South Korea, Russia, the USA, or Singapore), can prove useful, but must not be simply copied and applied to the current circumstances in Serbia. The principle of complexity and vigorousness in assessing risk and combating the pandemic would further involve simultaneous collection, monitoring, and interpretation of data in terms of economic, psychological, social, and other societal effects, in addition to analyzing statistical indicators of health and taking medical measures to save every life.

Analysis of the current state and scenario in Serbia's fight against COVID-19 has generated several questions: What is the best solution based on all currently available data; is the 20th century-based epidemiology and the manner of handling previous epidemics (the plague, Spanish flu, smallpox, anthrax, swine influenza, and avian influenza) really the best way to handle a virus with a mortality rate of about 1%; does isolation or mandatory hospitalization of asymptomatic cases make sense; how does a curfew contribute to the prevention of virus spread; and does the fear caused by insufficient knowledge about the virus justify all the measures taken?

It is reasonable to accept that the issue can be resolved using previously successful strategies. Alternatively, simple solutions can be offered to appease the populace or more drastic measures can be taken as if dealing with an extremely infectious and dangerous disease. Nevertheless, it is difficult to provide definite answers to the above questions that could validate simple strategies. First of all, decision-makers have to consider the assessment of the type and level of risk to which people are exposed as individuals, a group, or the entire nation. In addition, successful strategies and individual measures rely on the consensus across all levels and careful involvement of all stakeholders. Ultimately, the question is whether citizens should be forced to behave in a specific way or whether they should be allowed to choose such behavior, either because they trust the authority that imposes it or because they personally think it makes sense. It is well known that fear of repression can be a powerful tool for ensuring social obedience, but the patterns of behavior established this way cannot be sustained in the long run. If a single solution is implemented with strict persistence to minimize risk and avoid fatalities, individual solution strategies may emerge based on subjective risk assessment, which would, in turn, disrupt the established social structure. Likewise, the decision-makers have to take the time factor of the crisis and risk into account to be able to vigorously and flexibly adapt measures and solutions.

In times of collective stress and societal crisis, the characteristics of national culture should not be disregarded. Some individuals and communities have a highly pronounced need to avoid danger and uncertainty, so during a crisis they tend to adhere to their rigid beliefs and behavior, to be less tolerant toward innovations, and to feel an emotional need for strict rules. Under the current circumstances, such a trait of national culture has proven favorable for strategies that rely on enforcement, obedience, and preservation of order and control in some countries (e.g., South Korea, China, or Germany), whereas in other countries (e.g., Italy, Spain, France, and Serbia), it was difficult to implement the social measures of control and enforcement. It is true that specific regions and social strata were more ready to act obediently and to accept control, while others displayed a more liberal attitude and broke the rules of conduct during a state of emergency. All this ultimately boils down to the characteristics of risk management, the level of psychosocial stress, and the individual and social risk assessment. In new and unfamiliar situations, it is always beneficial to use well-established knowledge and skills, especially when they are certain to produce favorable results; however, in cases involving a high degree of uncertainty, anxiety, and stress, effective risk management is based on the preparedness to do something that others might find impossible.

4. Conclusions

At the turn of the 21st century, the balance of caring for human well-being and the quality of life shifted, with safety issues becoming more important, so risk assessment became a legal obligation when conducting numerous activities in many countries. Beck described the efforts that the stakeholders – governments, corporations, and others – expended to manage risk perception and risk management policies (Beck, 1992). If the natural disasters are accompanied by technological and anthropogenic crises and disasters, such as climate change, urban overpopulation, drinking water shortage in many parts of the world, and the risk of new diseases and epidemics like the ongoing COVID-19 pandemic which had destructive effects on every aspect of people's lives, then it is reasonable to assume that the remainder of the 21st century will be ridden with ever-emerging crises having ever more serious consequences, while the mechanisms in place used for prevention, preparedness, control, and response will fail to keep up. All the aforementioned factors impose an urgent need to establish new mechanisms of risk control both globally and nationally, as the crisis caused by the COVID-19 pandemic is still ongoing.

Conflicts of interest

The authors declare no conflicts of interest.

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Authors' contributions

All authors participated in the preparation and writing of the paper and meet all the criteria for authorship. Conceptualization: Milanko Čabarkapa and Teodora Safiye; writing – original draft preparation: Teodora Safiye and Milanko Čabarkapa; writing – review and editing:

Medo Gutić; visualization: Milanko Čabarkapa, Teodora Safiye and Medo Gutić; and supervision: Medo Gutić. All authors have read and agreed to the published version of the manuscript.

Ethical approval

Not applicable as this paper does not involve human subjects.

Availability of supporting data

Data utilized to this paper are from secondary sources and available to the public.

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RESEARCH ARTICLE

Accuracy of mother's reporting on child immunization in Yemen between 2012 and 2013

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Abstract

There is a complex interaction between infectious diseases and child nutritional status as infections usually entail some nutritional setback and vice versa. Therefore, vaccination against childhood infectious diseases is an important preventive measure against malnutrition itself, playing a key role in reducing child mortality. However, whereas referring to vaccination coverage it is crucial to have it clear how this coverage is measured, once it may vary considerably depending on the source of information. While child vaccination status is obtained from medical records in developed countries, in developing countries, they are mostly taken from vaccination cards and/or mothers' reports. Nevertheless, some researchers have come to diverse conclusions in terms of the accuracy of parents' reports. Based on a rich longitudinal household survey available for Yemen collected in 2012 – 2013, we find that although mothers' reports should not be discarded when estimating vaccination coverage (otherwise, coverage would be extremely overestimated), this information should be used with caution.

Keywords: Reporting accuracy; Vaccination coverage; Child immunization; Children; Maternal recall; Yemen; Yemen National Social Protection Monitoring Survey

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1. Introduction

There is a complex interaction between infectious diseases and child nutritional status in the sense that infections usually entail some nutritional setback and vice versa. Therefore, vaccination against the main childhood infectious diseases may be an important preventive measure against malnutrition itself (CSO, PAPCHILD, and MI, 1994). Reducing child mortality is defined by the United Nations (UN) as the fourth millennium development goal, and vaccination plays a key determinant to achieve this goal. Since 1974, the Expanded Program on Immunization (EPI) and the substantially increased vaccination coverage in many countries have saved the lives of millions of children. However, while referring to vaccination coverage, it is crucial to have it clear how this coverage is measured, as it may vary considerably depending on the way vaccination status is determined (Langsten and Hill, 1998).

Since 1998, the World Health Organization (WHO), United Nations Children's Fund (UNICEF), and selected ministries of health have developed the Joint Reporting Form (JRF) by consensus. The JRF is a standard questionnaire that was sent to all member states to disseminate consistent data on immunization system performance. The information collected in the JRF serves as a critical resource for tracking implementation of the Global Vaccine Action Plan (GVAP). These initiatives work as key frameworks to guide immunization strategies at global and regional levels and progress reports on GVAP (WHO, 2017).

While monitoring vaccination surveillance, there are two sources of vaccination coverage information, administrative records and specific surveys, which can be used to fill the gaps in administrative data. In this paper, we deal with data from specific vaccination surveys, which usually collect vaccination information through parents' recall and/or vaccination cards. Using the Yemen National Social Protection Monitoring Survey (NSPMS) data, the aim of the paper was to examine the level of accuracy of mothers' recalls on their children's vaccination histories. We did this by comparing mothers' reporting information in one round with the vaccination card shown in a subsequent round.

The usage of parents' recall and card information when analyzing vaccination coverage is widespread in the literature. Nevertheless, some researchers have questioned the validity of parents' recall of their children's vaccination. Although the literature indicates either over- or underestimation in parental recall, some researchers still conclude that including it in vaccination coverage yields more accurate estimates (Ndirangu, Bland, Barnighausen, *et al*., 2011; Brown, Monasch, Bicego, *et al*., 2002; Suarez, Simpson, and Smith, 1997; Langsten and Hill, 1998). However, there are also papers questioning parents' recall validity (Murray, Shengelia, Gupta, *et al*., 2003), and others even strongly do not recommend the use of parents' reports once they introduce recall bias (Valadez and Weld, 1992; Bolton, Holt, Ross, *et al*., 1998).

Vaccination coverage in Yemen has significantly improved between the nineties and mid-2000s, but there is still a long way to go for the country to reach vaccination coverage levels recommended by the WHO. For instance, 47% of children aged 12 – 23 months had received Diphtheria-Pertussis-Tetanus (DPT) vaccines in 1991 – 1992 (CSO, PAPCHILD, and MI, 1994), while 60% of them had received pentavalent in 2006 (MPHP and UNICEF, 2008). Since then, coverage has remained at the same level for some vaccines between 2006 and 2013, whereas for others, coverage has increased in the period. Aiming at boosting coverage levels, vaccination campaigns have been a constant effort in the country. In 2012 and 2013, there were nine polio vaccination campaigns. This may be the reason why polio vaccination coverage has significantly increased in Yemen, rising from 60% in 2006 (MPHP and UNICEF, 2008) to 74% in 2013 (International Policy Centre for Inclusive Growth [IPC-IG] and UNICEF, 2014).

It is worth highlighting the importance of mothers' reports in this context, as during most campaigns, vaccination may not be recorded on health cards so that survey enumerators can only obtain such information by parental recall (Langsten and Hill, 1998). Thus, it is very important to analyze the accuracy of mothers' reports. This is because on the one hand, if mothers' recall leads to coverage overestimation, children may be put at risk, as health workers may fail to vaccinate children who still need additional doses. On the other hand, if a mother's report leads to coverage underestimation, this may cause a waste of resources by vaccinating children already vaccinated (Valadez and Weld, 1992).

1.1. The history of Yemen's vaccination initiatives

Vaccination coverage in Yemen was very low until the 1980s. In the most favorable estimates, at most 10% of children were vaccinated (the Demographic and Health Surveys - DHS -1992 report). According to the WHO (1988), by the end of the 1980s, Yemen reported at least ten cases of poliomyelitis per year, and polio vaccination coverage was still lower than 30% (WHO, 1988). In 1990, with the vaccination for all campaign announced by the WHO, Yemen adopted national strategies for increasing vaccination coverage, reaching 80% of children vaccinated for most vaccines. However, before the mid-nineties, there was another relapse in vaccination coverage.

Yemen's vaccination coverage also suffered a noticeable drop after the mid-1990s because of a substantial fall in donor support starting in 1990 and a civil unrest taking place in 1994 (WHO and UNICEF, 2008). In 2005, it was not yet maintaining a consistent upward trend: having improved coverage levels in the early 2000s, it dropped again in mid-2003 (because of another poliovirus outbreak as a result of the spread of the virus from northern Nigeria) and recovered in 2004 (UNICEF, 2005; WHO, 2006). In 2005, Yemen still had active transmission of the imported virus, with 478 cases of children infected with poliomyelitis in the country. In that year, there were 6 national immunization days (WHO, 2006). The National Millennium Development Goals Report in 2010 pointed out that the national campaigns against polio carried out in Yemen in 2009 eliminated the disease in the country. Despite this, new cases were reported between 2011

and 2012. Similar results were found for measles, where campaigns showed very positive results in controlling the spread of the disease in the period between 2006 and 2009.

As can be noted, efforts to increase national vaccination coverage against the diseases covered by the EPI are part of a long-standing strategy to accelerate the reduction of child mortality in Yemen. According to UNICEF Yemen, several national and subnational vaccination campaigns took place in 2012 and 2013. The most recent vaccination campaigns are especially important when analyzing the youngest children – aged <2 years old who should have taken all vaccines by the time they were 1 year old (UNICEF and IPC-IG, 2014).

2. Data and Methods

2.1. The dataset

The NSPMS is the only nationally representative longitudinal household survey available for Yemen where members of 6397 households were interviewed on a quarterly basis during a 12-month period between October 2012 and September 2013. During the four rounds of the NSPMS, caretakers were asked to show the vaccination card for every child under age five. The interviewers copied the vaccination information from the card onto the NSPMS questionnaire. If there was no vaccination card, the mother was asked to recall whether the child had received each of the vaccines, and in the case of multiple doses, interviewers also asked how many times the vaccine was given. Given the longitudinal nature of the NSPMS, households were visited 4 times over a 12-month period. These sequential visits allowed the survey to improve the information on children's vaccination histories, as they increased the likelihood of having access to vaccination cards and to find better informed interviewees on the children's vaccination histories.

This paper focuses on Bacillus Calmette-Guérin (BCG) and the third dose of polio vaccines (polio 3rd). Typically, indicators of children's vaccination coverage consider children aged 12 – 23 months as their target population. This is important because one would expect the memory error of mothers to be greatly reduced when considering the immunization schedule of very young children.

There were 1369 children aged 12 – 23 months in round 3 of the NSPMS data. It is important to emphasize that our sample comprises children aged 12 – 23 months in round 3 whose card was seen in this round and whose information on vaccines in round 1 was taken through the mother's report. In addition, concerning the dates of vaccination obtained in round 3, we only kept children with dates of vaccination that was consistent within this round. It is important to emphasize that for a particular child, their information on vaccines comes from different sources. For instance, it is possible for a child to have a consistent date at which BCG vaccine was taken but having no record on the card about third dose of polio and their mother reported them as not vaccinated against polio. This means that for the same child, it is possible to have information from either the vaccination card (through a readable or not readable date of vaccination) or the mother's report depending on the vaccine to be considered. In other words, card availability information varies obviously from child to child, but more than that, it varies from vaccine to vaccine even when considering the same child. Because of this type of vaccination history, the number of sampled children varies depending on the vaccine that is being used to analyze the accuracy of the mother's report.

2.2. Methods

The methodology for verifying the accuracy of mother's reports on child vaccination is adapted from Langsten and Hill (1998), and it is merely descriptive. First, we excluded from the analysis children with inconsistencies in the dates of vaccination copied from their vaccination card in round 3. That is, we excluded children whose (1) dates of vaccination were before their own birth and (2) vaccination dates in one round were after the date of the interview of that round. In addition, some children also lack information concerning their vaccination status, which means either "mother doesn't know," "mother doesn't remember" or "children not vaccinated." For the majority of children with missing information, the interviewer had seen their vaccination card, and we assumed that these children were not vaccinated according to the card.

Then, we analyzed the possible sources of disagreement between the mother's report in round 1 (R1) and the card information in round 3 (R3). Since data collection improved over time, it is important to mention that our definition of consistency criteria on mother's recall considers the round 3 information as our benchmark. After checking the consistency of the mother's report, the following metrics were calculated: accuracy, sensitivity, specificity, and predictive values. These measures are commonly used in research to validate and compare instruments (Selimuzzaman, Ullah, and Haque, 2008; Miles, Ryman, Dietz, *et al.*, 2013).

The accuracy of mothers' reporting corresponds to the percentage of children whose mothers reporting information matched the information registered in their vaccination card: (TP + TN)/(TP+FP+FN+TN). Sensitivity is the percentage of vaccinated children who

were reported by their mother as vaccinated: TP/(TP+FN). Specificity is defined as the percentage of nonvaccinated children who were reported by their mothers as not vaccinated: TN/(FP+TN). The positive predictive value (PPV) is the percentage of children reported as vaccinated who were actually vaccinated: TP/(TP+FP). Finally, the negative predictive value (NPV) is the percentage of children reported as not vaccinated who were actually not vaccinated: TN/(FN+TN). In each of these equations, the acronyms mean the following: TP=true positives, TN=true negatives, FP=false positives, and FN=false negatives. The definition of true or false (positives or negatives) refers to the consistency between information on vaccination cards and mother's recall, taking into account that the former is our gold standard.

Based on these measures of agreement, we evaluated the level and pattern of accuracy of mothers' recall on their children's vaccination histories by comparing mother's reporting information in round 1 with the vaccination card shown in round 3. It is important to emphasize that validity refers to the degree to which the test or an estimate based on a test is able to determine the true value of what is being measured (in this case, vaccine coverage). In this sense, sensitivity and specificity (and therefore the accuracy itself) are properties inherent to the criterion of the evaluated test and do not vary except by technical error. Predictive values, however, depend on the prevalence of the phenomenon in the study population. The PPV increases with prevalence, while the NPV decreases. Thus, when the phenomenon is rare, the PPV is low, since most of the positive information results from mothers of unvaccinated children, representing false positive results. On the other hand, NPV is high at low prevalence (Fletcher, Fletcher, and Fletcher, 2014).

3. Results

3.1. Vaccination coverage by source of information: mother's report versus vaccination card

Figure 1 shows the percentage of vaccinated children – against BCG and polio (third dose), fully vaccinated (had received BCG, measles and third dose of polio and pentavalent), undervaccinated (had not received at least one of them) and even not vaccinated at all – among those whose information on all vaccines was available through their mothers' reports and the percentage of vaccinated children among those whose information on all vaccines was available on their vaccination cards. It is important to emphasize two features of this figure. First, it only includes children with available information on all vaccines (either from mother's report or vaccination card). Although having all children with complete information on their vaccination status would be the ideal situation, only 62% of children aged 12 – 23 months who have vaccination cards show information on all vaccines taken, and the percentage of children aged 12 – 23 months who have a vaccination card is as low as 42%. Second, it is important to highlight that the sample of children used in this table does not exactly correspond to the sample of children used in our empirical exercise. The idea of Figure 1 is giving a more general idea of how the estimates in vaccination coverage may vary depending on the source of vaccination information (whether it is mother's report or vaccination card).

The most striking aspect of these numbers is the significantly higher percentage of vaccinated children among those with vaccination cards in comparison to those whose mothers reported their vaccination status. For instance, approximately 83% of 557 children aged 12

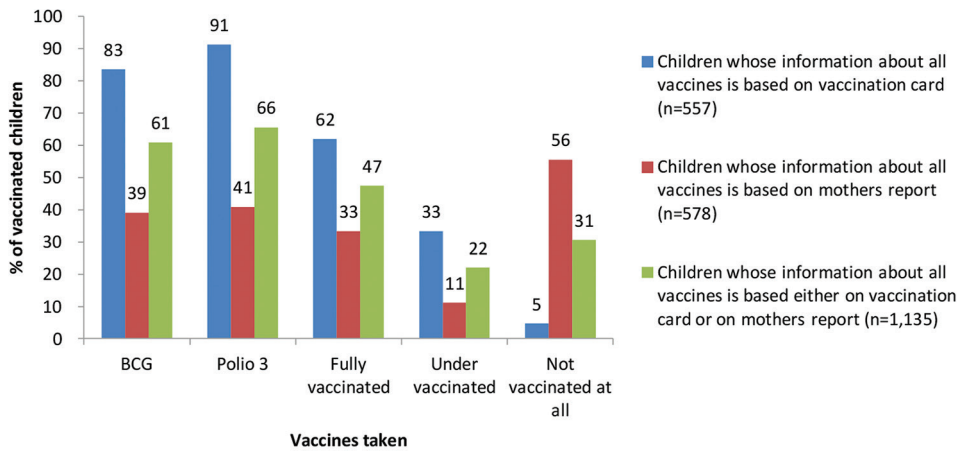

Figure 1. Percentage of vaccinated children among those aged 12 – 23 months in round 3 with information available on all vaccines taken according to the source of information – Yemen, 2013. Source: IPC-IG and UNICEF (2014), round 3.

– 23 months with vaccination cards available have received BCG. Among the 578 children whose mothers are their source of information, only 39% have received it. Whether we analyze children who have never taken any vaccine, only 5% of the children with vaccination cards fall in this category against 56% of children whose information comes from the mother's report. This is an indication of two possibilities that may be complementary to each other: (1) mother's recall of child vaccination is very incomplete in the sense that it probably underestimates children's vaccination status, and (2) vaccination cards are a key source of children's vaccination history.

3.2. The accuracy of mother's reporting on child vaccination status

We tested mother's precision while reporting their children's vaccination status regarding two main issues: (1) The overall quality of mother's reports based on both mother's reporting information and subsequent vaccination card information and (2) the direction of mothers misreporting, which indicates whether mother's report mostly leads to underestimation or overestimation of children's vaccination coverage.

3.2.1. Overall quality of mothers' reports

As proposed by Miles *et al.* (2013), we specified ≤80% as being a poor validity of the instrument/source of information (in this case, mother's report on vaccination status of children aged 12 – 23 months). For both vaccines, we found low agreement between the mother's reports in R1 and vaccination cards in R3, with accuracies of 67 and 71% for BCG and polio 3rd, respectively.

3.2.2. The direction of mothers' misreporting

Table 1 shows the sensitivity, specificity, PPV, and NPV while comparing the mother's report at R1 with the vaccination card at R3 (the latter being our gold standard). Before analyzing the results, we highlight that due to our sample restrictions, the sampled children are in better socioeconomic conditions compared to the others although, on average, both groups of children (those included or those not in the sample) do not have adequate living conditions as access to health facilities, water, sanitation, education, and food security are still scarce in a country where poverty rates were estimated at 45% in 2012 (IPC-IG and UNICEF, 2014). Although these conditions can affect both groups homogeneously, this possibility cannot be tested, which introduces a limitation to the study.

The sensitivity values were higher than the specificity, and the PPVs were higher than the NPVs for both vaccines. When comparing the type of vaccine, we easily see that there was relatively little change in sensitivity between them. Taking into account the cutoff of ≤80% as a low value, sensitivity was low for both vaccines, with BCG showing the lowest value (70%). Considering the specificity, the values were even worse for both types of vaccines, and BCG again presented the lowest value (56%). PPVs were high for polio 3rd and BCG (88 and 84%, respectively). In contrast, NPVs were low for both (38 and 36%).

These results indicate an acceptable effectiveness of the instrument (mother's report) while estimating vaccination coverage. However, some features should be highlighted. According to the results presented in Table 1, mother's information is more reliable when predicting children who were actually vaccinated, but it is more problematic when predicting those who were not vaccinated. This means that mothers usually underestimate the number of vaccines taken by their children in Yemen, which in turn may underestimate children's vaccination coverage, as for most children, the main source of information on their vaccination schedule is their mother (59% of children aged 12 – 23 months in Yemen does not have a vaccination card available). In fact, as we assumed that the gold standard for the assessment of coverage was the vaccination card, it is estimated that the vaccination coverage is 76.92% and 79.51%, respectively, for BCG and polio 3rd in our sample of children. The estimated coverage for the same sample based on mother's recall is more than 10 percentage points lower than that based on the vaccination card (64.10% and 66.34%, respectively).

4. Discussion

The previous sources of vaccination data in Yemen are the 1991 – 1992 and 1997 DHSs and the 2006 MICS, both offering access to cross-sectional data. Nevertheless, while adequately analyzing the accuracy of parents' recall, it is crucial to have access to longitudinal information. This is because in a household survey interview, caretakers are usually first asked to show the vaccination card of each child, and only in case they do not have it are they asked to recall whether the child had received each of the doses/vaccines. In this way, in cross-sectional data, the researcher will only have access to one source of information, either the vaccination card or parents' report. Based on the NSPMS longitudinal survey, it was possible to test the accuracy of mother's reports in Yemen by comparing mother's information at R1 with the information collected at R3 (6 months later).

It is important to highlight that although we also tested mothers' accuracy in a sample including all children aged 12 – 59 months, we found that memory errors increased significantly with age. In this sense, to minimize miscalculating vaccination coverage indicators

Table 1. Measures of agreement between vaccination card ("gold standard") and mother's report and vaccination coverage according to both sources of information – Yemen, 2013.

Agreement of instruments/sources of information	Children aged 12 – 23 months		
	Vaccinated children based on Card info at R3?		
	Yes	No	Total
BCG			
Vaccinated children based on mother's report at R1?			
Yes	105	20	125
No	45	25	70
Total	150	45	195
Indicators of Agreement			
Accuracy		66.67%	
Sensitivity		70.00%	
Specificity		55.56%	
PPV		84.00%	
NPV		35.71%	
Prevalence (vaccination coverage)			
Gold standard		76.92%	
Mother´s report		64.10%	
Polio 3rd			
Vaccinated children based on mother's report at R1			
Yes	120	16	136
No	43	26	69
Total	163	42	205
Indicators of Agreement			
Accuracy		71.22%	
Sensitivity		73.62%	
Specificity		61.90%	
PPV		88.24%	
NPV		37.68%	
Prevalence (vaccination coverage)			
Gold standard		79.51%	
Mother's report		66.34%	

Source: IPC-IG and UNICEF (2014), rounds 1 and 3.

in developing countries (where vaccination coverage estimates often depend on parents' information), we recommend it to be mainly estimated for children aged 12 – 23 months. The results for these children showed that the sensitivity and specificity had moderate values, except for BCG. This may be because this vaccine is given at birth, henceforth more subjected to parent memory errors when they are questioned, despite its typical scar. In this sense, an analysis that includes the temporal distance between the vaccination and the interview, although being an interesting complementary analysis, is beyond the scope of this study. Regarding the predictive values, while the PPVs were high, the NPVs were very low for both vaccines.

There is no consensus about the most appropriate thresholds to determine whether an instrument is valid to estimate vaccination coverage. We used a more restrictive criterion proposed by Miles *et al.* (2013), in which each measure of validity must be >80%. For service planning within the health sector, it is essential that the instruments used to evaluate the coverage of services present high levels of sensitivity and specificity. These features enhance the ability of policymakers to capture the actual distribution of a certain condition in

the population, namely, children's vaccination coverage, in the case of this study.

From the clinical point of view, specificity is a more important characteristic, since it refers to a correct diagnosis for which we do not wish to make a mistake (i.e., to assign a positive diagnosis when it is not true). However, from the point of view of public health, actions are expected to be as sensitive as possible since it is more important to ensure high coverage for disease screening or preventive action coverage. Thus, in the case of vaccination, the higher the sensitivity, the better it is for the analysis of vaccine coverage because even though there is an overestimation of the coverage, it is assumed that the target population is covered by the action.

The moderate sensitivity associated with a high PPV indicated that the mother's report is highly accurate when identifying the proportion of children who were vaccinated in the Yemen population. However, the low NPVs indicated that many children may be misclassified as not vaccinated when they have actually been vaccinated. However, it is important to stress that although mothers' reports should not be discarded while estimating vaccination coverage in Yemen (otherwise, coverage would be extremely overestimated if only estimated based on vaccination cards), this information should be used with caution. From our results, we find that considering mother's information in estimating vaccination coverage in Yemen tends to underestimate this indicator (for instance, BCG vaccination coverage according to vaccination card is 77%, if based on mother's report, coverage decreases to 64%). If, on the one hand, this finding may cause certain relief as the real situation of child vaccination in Yemen may actually be better than the one based on the estimated coverage; on the other hand, there seems to be a waste of the already scarce resources of the country. With this analysis alone, we are not able to estimate a correction factor for an official vaccination coverage estimate (based on mother's report and/or vaccination card), as this factor would be strongly dependent on the prevalence of vaccination in Yemen, and we have a sample of those with (consistent) information. In addition, it would be important to analyze the characteristics of the families of children with vaccination card information compared to those with only mother reports. Further analysis is required to move forward at this point.

There are successful national immunization programs (NIPs) implemented in developing countries that could help other countries that still present low child vaccination coverage and limited resources – such as Yemen – rethink their immunization systems. Brazil has been considered an international case of success: eradicated smallpox in 1971, instituted the NIP in 1973, and held its first national campaign against polio in 1980. Despite all the advances, until the mid-1990s, the records of vaccination were not satisfactory. By the end of the 1980s, with the establishment of the Brazilian Unified Health System, a movement of decentralization was initiated that placed the municipality as the direct executor of health actions; among them, vaccination was included. It is worth mentioning that this decentralization in the actions is based on the integration between three levels — municipal, state and federal. With a national vaccination registration system, these three levels together discuss norms, definitions, goals, and results, providing the continued modernization of its infrastructure and operation. In this scenario, the NIP has ensured the provision of safe and effective vaccines with extremely high vaccination coverage for children (Silva Jr, 2013).

However, for the Brazilian NIP to be such a success, in addition to the program being part of the WHO's program, the Brazilian government also had the support of children's rights organizations such as UNICEF and the Pan-American Health Organization (Ministério da Saúde, 2014). In addition, the Brazilian Ministry of Health defined the NIP as one of its priorities, allocating resources to it in the annual budget in a separate item and not subjected to budget cutting as per the Budget Guidelines Law of 2011 (Domingues, Teixeira, and Carvalho, 2012). To reinforce the importance of maintaining high vaccination coverage among children in Brazil, the Ministry of Health in 2004 issued an ordinance making it mandatory to present children's vaccination cards in case of child enrollment in school, parent job hiring and receipt of social benefits. In case parents do not have the card in hand, the institution gives 2 months for them to present children's updated vaccination card. This shows the importance of a vaccination card, which should be reinforced in Yemen not only among families but also among health professionals. Vaccination cards can enhance health workers' ability to make decisions and empower caregivers on their child healthcare in addition to supporting public health monitoring (Brown, 2012).

According to Yemen's Planning Strategy for Immunization Service Delivery in a Catchment Area (MPHP, 2005), every year, each health facility is responsible for conducting a house-to-house census of the population eligible for immunization in their catchment area. Among the purposes of the annual house-to-house census are (1) verifying the immunization status of children aged <1 year and women of childbearing age; (2) providing immunizations to eligible children and women and providing them with immunization cards; (3) updating the records of immunizations given; and (4)

educating men and women on the safety and importance of immunizations based on local needs. Despite all these efforts in guaranteeing population access to immunization cards at the facility level, families seem to not understand the importance of keeping vaccination cards at hand. This becomes clearer when using data on vaccination from Yemen's household surveys, as much of the information relies on parents' reports instead of vaccination cards.

5. Conclusion

Based on the sample of children used in our analysis, we found that considering mothers' reporting information in estimating vaccination coverage in Yemen tends to underestimate this indicator. Although this finding may cause relief, as the real situation of child vaccination in Yemen may actually be somewhat better than that based on the estimated coverage, this also means a waste of the already scarce resources of the country. An important fact to be mentioned is the considerable percentage of children who are born at home without the presence of skilled health personnel during childbirth in Yemen. Approximately 73% of women did not deliver births at a health facility in 2013. Moreover, only 14% of women delivering at home were attended by skilled health personnel during childbirth (IPC-IG and UNICEF, 2014). This fact is especially important when taking into account BCG vaccine (and first dose of polio), which is given at birth.

It is important to enhance families' and health workers' awareness of the importance of keeping children's vaccination schedules up-to-date. Maybe the fact that mothers make more mistakes answering that their child was not vaccinated when they were actually vaccinated indicates that mothers find vaccination somehow important – as if they do not remember whether their child was vaccinated or not, they may think it is better saying the child was not immunized so that they can be (re)vaccinated anyway. However, as highlighted earlier, this means that the resources used for child immunization in Yemen could be used more efficiently. Finally, while it is essential to improve the population's understanding in terms of the importance of the vaccination card, the centralization of the registration system and the continuous recording of children's vaccination can be seen as a recommendation for improving the management of childhood vaccination at the national level.

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Conflict of Interest

No conflicts of interest were reported by the authors.

Authors' Contributions

Conceived and designed: Laetícia Rodrigues De Souza. Review of literature: Laetícia Rodrigues De Souza and Luciana Correia Alves. Contributed to tools/materials/data collection: Laetícia Rodrigues De Souza, Luciana Correia Alves and Raphael Mendonça Guimarães. Analyzed the data: Laetícia Rodrigues De Souza, Luciana Correia Alves and Raphael Mendonça Guimarães. Drafted and wrote the manuscript: Laetícia Rodrigues De Souza and Luciana Correia Alves.

Ethical Approval

The human data used in our research are a publicly available household survey dataset that can be downloaded from the webpage http://nspms-yemen.ipc-undp.org/.

Availability of Supporting Data

The NSPMS dataset is in open access on the webpage dedicated to the UNICEF and IPC-IG joint project (http://nspms-yemen.ipc-undp.org/).

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ACCSCIENCE PUBLISHING

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RESEARCH ARTICLE

Japan's death-laden society: Five areas of prospective policy challenges

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Abstract

Today, Japan stands as the world's leading super-aged society. In the coming decade, preceding the rest of the aging globe, the country will phase into the next demographic stage called a "death-laden" society. Due in part to the aging and prospective mortality of the country's two major baby boom generations along with a projected decline in the number of the working age population, Japan will be laden with ballooning deaths from old age from 2030 onward for several decades to come. Only in recent years have researchers started paying attention to this demographic prospect, and to date, little study has been done to systematically examine how the coming of a death-laden society may affect the health, well-being, and comfort of those in advanced age in the country. This paper aims to contribute to the newly emerging body of literature on this subject by exploring, based mainly on findings from expert interviews, five key areas of policy challenges with which Japan's death-laden society will likely contend. These areas include: (1) *shortages in basic medical resources for the dying*; (2) *mounting public burden of disease*; (3) *potential prevalence of 'lonely deaths' among those in advanced age*; (4) *urgency to facilitate national discussions on end-of-life options*; and (5) *crematorium shortages and their cultural impact*. The future research is called for to help mitigate the impact of a death-laden society not only for Japan but also for other countries that may follow Japan's demographic path in the conceivable future.

Keywords: Population aging; Japan; Death-laden society; Death and dying; Policy

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1. Introduction: The coming of Japan's 'death-laden' society

Over the past several decades, Japan has led the rest of the world in population aging. Preceding the rest of the aging globe, in 2007, Japan grew to be a "super-aged" society, a stage of population aging in which those aged 65 or older account for at least 21% of the total population (Higo and Klassen, 2016). By October 2021, the figure had reached 29.1%, a much higher rate than that of any other super-aged societies in the world. The dependency ratio of older people to those of working age – those aged 65 and over to those aged between 20 and 64, including both men and women – have also significantly increased over the past two decades; the ratio jumped from 28.7 in 2002 to 55.2 in 2021 (United Nations, 2022). The ratio for Japan in 2021 stood substantially higher than for any other country, suggesting that the burden of population aging on the younger population is greater for Japan than for any other part of the world. While

most of today's other super-aged societies are European countries including Italy, Portugal, Finland, and Greece, just in the coming few decades major economies in East and Southeast Asia – most notably, South Korea, China, Taiwan, and Singapore – will catch up with the rate of Japan (United Nations, 2019).

In recent years, a growing amount of literature has argued that just in the coming decade a super-aged Japan will transition into what is often referred to as a "death-laden" society (Fuji, 2018; Kurahara, 2020; Nagaoka, 2021). A death-laden society is characterized by a ballooning of deaths among its population, particularly of those in advanced age, and mounting challenges to the whole society, including the younger population, in its effort to protect individual members' wellbeing, comfort, and dignity in later life.

In this new stage of demographic shift, while remaining the world's leading super-aged society, Japan will experience a ballooning of deaths among its population, particularly of those in advanced age. During the second half of the 20th century, annually about 0.8 million people died on average, and that figure has rapidly increased to date. In 2030, the figure will reach about 1.6 million, which will remain through the 2060s. By contrast, the number of births has been steadily declining during this period of time in a nearly reverse fashion. The annual average births during the second half of the 20th century were about 1.6 million, and the projected figure for 2030 is about 0.8 million (United Nations, 2022). Nearly 80% of those deaths are projected to occur among those in their mid-70s and older (National Institute of Population and Social Security Research, 2022). This trend is driven in part by the prolonged longevity; the life expectancy at birth for both men and women jumped from 67.3 years in 1960 to 84.7 years by 2020, an almost 25% increase over the past five decades (Organization for Economic Co-operation and Development, 2022). Another factor behind the ballooning of deaths is the continuous aging and eventual deaths of the two major baby boom generations of the country's population. Together, these factors will result in rapid depopulation due mainly to a projected continuation of the decline or stagnation of childbirth (Higo, forthcoming). Hence, from 2030 onward for several decades to come, Japan will be a society that is characterized not only by the continuous aging of the population but also by being laden with a lasting trend of experiencing an unprecedented number of deaths in its older population. Much of the emerging body of the literature has focused on describing a death-laden society mostly from a demographic perspective or analyzing the causes and process of the demographic transition. To date, notably understudied has been the prospective consequences of this transition – how the coming of a death-laden society will challenge Japanese society, culture, and individuals including those in advanced age.

This paper aims to contribute to the growing body of the relevant literature by exploring, albeit preliminarily, main areas of challenges, particularly from a policy perspective, with which a death-laden Japan will likely contend at least for several decades to come. Drawing on survey data that are publicly available and original data gathered from a series of expert interviews, this paper outlines five areas of challenge that call for immediate policy responses to protect the well-being, comfort, and dignity of those in advanced age not only today but also in the future. The five areas of challenge include: (1) Shortages in basic medical resources for the dying; (2) mounting public burden of disease; (3) potential prevalence of "lonely deaths" among those in advanced age; (4) urgency to facilitate national discussions on end-of-life options; and (5) the crematorium shortage and their cultural impact. An overarching goal of this paper is to contribute to the policymaking not only of Japan but also of other countries that may follow Japan's demographic path to becoming death-laden societies in the conceivable future, those in East and South-east Asia in particular. The experience of Japan as the world's prospective forerunner of a death-laden society may offer a source of policy lessons that may help those countries prepare to make the future of their aging societies more sustainable for the coming generations.

2. Data and methods

The discussion in this paper is based on two sets of sources: The first set of sources is a review of the relevant literature, both academic and policy, and findings from a series of descriptive analyses of survey data that are publicly available. These sources were utilized mainly to describe the basic characteristics and trends of Japan's death-laden society. The second is a set of findings from original data gathered in recent years from expert interviews, a qualitative research method instrumental to gaining in-depth information about specific issues, typically emerging ones, that are not necessarily publicly recognized (Döringer, 2020). The expert interviews were carried out to access and gather the latest information and specialized knowledge related to policy challenges with which Japanese society will likely contend over the coming decades particularly in the context of its continuous population aging. Together, these two sets of sources aim to empirically explore and discuss the policy prospects of Japan as the world's first death-laden society.

The expert interviews were conducted over the time span from February 2020 to October 2021 in Tokyo, Osaka,

and Nagoya. These locations were chosen because these are the most populous cities in the country and may also help capture some potential regional differences in interview participants' perspectives. A total of 24 experts participated in the interviews, including government officials, medical and health-care practitioners, representatives of medical institutions, and researchers affiliated with universities and think-tank organizations. Initially, three of the participants were invited to be interviewed due to their organizations' publicized concerns specifically regarding Japan's prospective transition into a death-laden society. Their concerns and discussions were publicly expressed in their whitepapers, newsletters, and online articles, among others. Then, a snowball sampling method was utilized to recruit the rest of the participants.

The interviews were designed to be in-depth and semi-structured and conducted in a one-on-one manner. Each interview followed a set of prepared questions centering around participants' accounts of their current concerns and challenges in pursuing their professional missions specifically in the context of the country's population aging and how those concerns and challenges will likely evolve in the conceivable future. In addition to exploring the participants' views of these prepared questions, the interviews also left some flexibility for unexpected themes to emerge. Due to the travel restrictions under the country's declaration of a state of emergency, nearly half of the interviews were carried out remotely using various ICT software programs including Zoom and Webex. All the interviews were voice-recorded and anonymously administered. The data were analyzed with a series of thematic coding and analyses to categorize specific concerns and challenges conveyed by the interview participants into broader themes. As a result of the thematic analysis, five themes in the data emerged, each of which is mutually related in reality and yet conceptually distinguishable enough to be discussed as a unique area of policy challenge in itself. In what follows, these five themes are discussed as five key areas of policy challenges that Japanese society will likely be urged to address on the verge of transitioning into a death-laden society.

3. Results and discussions

3.1. Shortages in basic medical resources for the dying

The first area of policy challenge for a death-laden Japan is prospective shortages in clinical doctors and hospital beds, basic medical resources necessary for those spending their past days of life at conventional medical institutions such as hospitals, due to the ballooning deaths in the country's population. Clinical doctors have been in short in Japan since well before reaching the verge of becoming a death-laden society. This has been a publicly recognized issue since the mid-1950s, particularly in rural areas and on remote islands (Matsumoto, Kashima, Owaki, *et al.*, 2019). In addition, over roughly the past two decades, an increasing number of semi-rural areas and mid-sized cities across the country have come to face the same issue due in part to the migration of younger populations to larger cities, including those working in medical fields (Takata, Nagata, Nogawa, *et al.*, 2011). Dying at hospitals typically requires that doctors provide medical treatment, care, and support just as they do when treating injuries and illnesses of patients aiming to recover. In many cases, intensive care, extensive treatments, and use of advanced technologies are called for on behalf of those patients whom the doctors know are going to die at their hospitals (Interview data, Tokyo, March 22, 2020). It is highly likely that the coming of a death-laden society will exacerbate the doctor shortages that the country has been suffering from for decades (Interview data, Tokyo, August 15, 2021).

A death-laden Japan will also run in short on hospital beds. This will be a major challenge as hospitals are the predominant place of death among the Japanese today. Figure 1 illustrates the composition of place of death among the citizens in 2020. In this year, about 68.3% of all deaths took place at hospitals. Those who died at home in the same year accounted for only about 15.7% of all deaths, and the corresponding figures for retirement homes and long-term care facilities were approximately 9 and 3%, respectively. While in recent years, the figure for homes has been on the rise, the majority of Japanese people – nearly 70% – die at hospitals today (Figure 1).

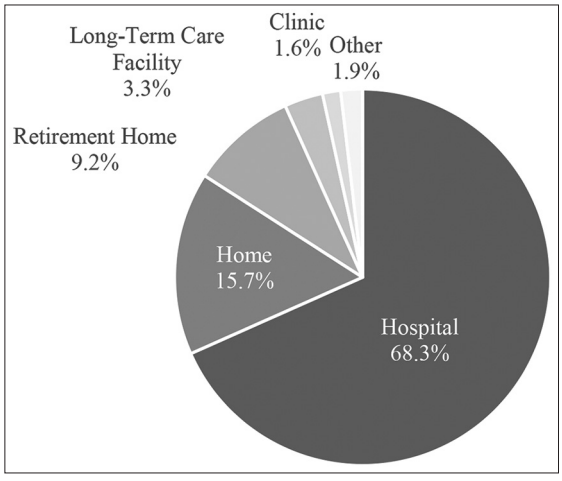

Figure 1. Composition of place of death, Japan, 2020 (%).
Source: Ministry of Health, Labor and Welfare (2022)

It is worth noting in this context that to date, the impact of the COVID-19 pandemic on this trend has remained unclear. Japan first declared a state of emergency on April 7, 2020, following the World Health Organization's declaration of a global pandemic on March 11, 2020. According to the Ministry of Health, Labor and Welfare (2022), the figure was 71.3% as of March 2018, roughly 2 years before the state of emergency, and 68.7% as of March 2021, about a year after the first declaration of the state of emergency. While no substantial change is observed during this period of time, one may argue, data of the coming few years are necessary to make a more accurate projection of the projected shortage in hospital beds.

Japan is unique among developed countries in this trend that hospitals are the primary place of deaths for the population. Since the early 1990s, an increasing number of countries in Europe and North America have come to promote not only "aging in place" but also "dying in place" in response to a growing demand from those of advanced age and those dying who call for respecting individual dignity, rights, and autonomy in choosing where to die (Cohen and Gott, 2015). Policy responses in those countries include expanding home care services and increasing the number of hospices for those citizens who desire to spend their past days at the places with which they feel most familiar and comfortable – typically their own homes or hospices in their communities, instead of hospitals (Gutiérrez-Sánchez, Gómez-García, Roselló, *et al.*, 2021). In the case of Japan, while medical communities have raised their voice to follow such overseas trends, no official policy measure has been announced to date, at least at the national level (Interview data, Osaka, April 8, 2021). If hospitals continue serving as the dominant place of death, a death-laden Japan will inevitably face a significant shortage in the conventional pace to die: Hospital beds. According to an estimate by the Research Institute of Economy, Trade and Industry (2018), as of 2019 about 882,000 people aged over 70 died in hospital beds, which consumed approximately 90% of the capacity of all hospital institutions across the country. In 2030, the number for the same age group is projected to reach nearly 1,460,000. Assuming that today's rate of hospital death will remain the same, by 2030, the annual shortage of hospital beds is projected to be for about 600,000 patients, and the number will only grow over the succeeding decades (Research Institute of Economy, Trade and Industry, 2018).

3.2. Mounting public burden of disease

Against the backdrop of global population aging, "burden of disease" – the impact of health-related challenges on various aspects of society, including the financial cost of care for older populations – has been a primary concern among most developed countries across the world (World Health Organization, 2008). Japan is no exception; as the world's leading super-aged society, the country has been contending with growing public health-care expenses for decades (Gilmour, Liao, Bilano, *et al.*, 2014). This burden of disease is therefore the second area of challenge for Japan in its transition to a death-laden society, with the financial costs projected to reach insolvency. This is the case particularly if no major change is made to the way in which people have conventionally relied on medical and health-care resources during the last stage of their lives (Interview data, Nagoya, September 28, 2020).

Figure 2 illustrates the trend of national health-care expenditures for medical care and long-term care from 2000 through a projected 2040. In the country's categorization scheme in the public expenditure, briefly put, medical care refers to that provided by hospitals and clinics, and long-term care to that offered by other institutions serving for older people, including homecare providers and hospices. In-home care is categorized into the former so long as it is provided by hospitals or clinics for medical purposes. Both medical and long-term care fall under the coverage of the National Healthcare Insurance and the country's universal health-care program, which is financed largely by the working-age population through their payroll taxes and premium contributions (Ministry of Health, Labor and Welfare, 2021). Especially over the past two decades, national expenditures on these areas have been rising rapidly; in 2000, the expenditure on medical care was 202.9 billion USD, and that on long-term care was 26.3 billion USD. By 2020, the figure for each area had increased to 328.7 and 116.9 billion USD, respectively. According to a recent projection made jointly by the Cabinet Office, the Ministry of Finance, and the Ministry of Health, Labor, and Welfare (2018), the figures for both areas will continue

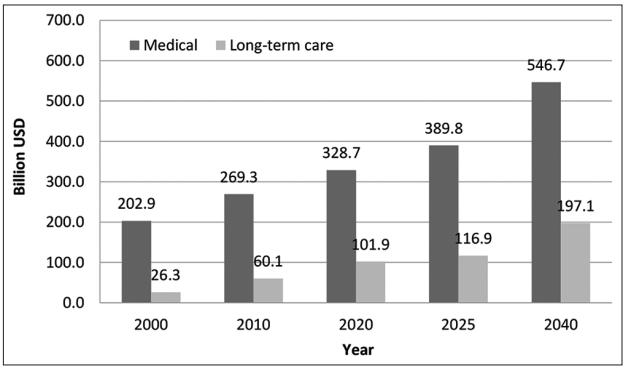

Figure 2. National health-care expenditures for medical care and long-term care, Japan, 2000-2040* (billion USD).
Source: Cabinet Office, Ministry of Finance, and Ministry of Health, Labor and Welfare (2018)

increasing in the future due to anticipated growth of health-care needs associated with the continuous aging of the population over the coming decades. By 2040, the expenditures are projected to jump to 546.7 for medical care and 197.1 billion USD for long-term care (Figure 2).

This projected public burden of disease is unsustainable and should the current framework to finance the expenditures be maintained in the era of a death-laden society (Interview data, Tokyo, 6 July 2021). As discussed above, the majority of people in Japan die at hospitals, for which significant financial resources are spent providing medical care for patients suffering terminal health conditions. Over the next decade, the country may see an increase in the number of people who die, or choose to die, at long-term care facilities as an alternative to the conventional, hospital deaths (Interview data, Tokyo, July 6, 2021). Whether dying at hospitals or long-term care facilities, however, the ballooning of deaths from old age will only contribute to pushing the burden of disease to insolvency. Simultaneously, this challenge will be further exacerbated by a continuing decline in the working age population who primarily finance the public health-care budget.

3.3. Potential prevalence of "lonely deaths" among those in advanced age

The third area of challenge is a possible adverse effect of promoting "dying in place" for those in advanced age in Japan who wish to die at home rather than in hospitals. Despite hospitals remaining the dominant place of death, the majority of older people in the country would actually prefer spending their final days at their homes. According to an opinion survey conducted in 2020 with a nationally representative sample of those aged 75 to 84, above 64% of them expressed their desire to die at their own homes. Moreover, over 85% of those preferring their homes reasoned that they considered that being at home would allow them to maintain a sense of self, comfort, and dignity up until the last moment of their lives (Nippon Foundation, 2021). Policymakers in the country might find it urgent to follow other countries in promoting "dying in place" both to accommodate such preference of today's older population and to help mitigate the prospective growth of public health expenditures.

However, pursuing this policy direction may create a potential adverse effect: A prevalence of what is often referred to as "lonely deaths" among those in advanced age. A "lonely death" refers to an incident when an older person living alone spends his final years living alone and then dying at home without care or attention from others, including his own relatives, after which the deceased body is discovered by an unacquainted neighbor weeks or months after his death (Kato, Shinfuku, Sartorius, *et al.*, 2017). Older people living alone in urban areas are likely to face a greater risk of lonely deaths than those in rural counterparts due mainly to a greater likelihood for the former to be childless and have weaker ties to the communities in which they reside (Interview data, Tokyo, July 19, 2021).

In the Tokyo area, for instance, incidents of "lonely deaths" have been rapidly surging in recent decades. Figure 3 illustrates the trend from 2002 through 2021 of the number of those aged 65 or older who died at home alone and were investigated by the Tokyo Medical Examiner's Office on the causes of death. One thousand two hundred and seventy-four people allegedly died "lonely deaths" in 2002, and the number jumped to 5258 by 2021, above a four-hold increase in two decades (Tokyo Medical Examiner's Office, 2022). To date, the data for the whole country are not readily available. The case of Tokyo, nonetheless, may suggest that a similar trend has been taking place in other major cities also, and that in the coming decades, this grim phenomenon will likely prevail among a larger share of older people across the country (Interview data, Tokyo, October 3, 2021).

Dying in place with a sense of comfort and dignity requires, in many cases, close care and quality support from informal caregivers – the dying person's spouse, children, or siblings in particular – especially during the person's last days (Brink, 2008). In Japan, however, over the past several decades, the number of older people who live alone has been steadily increasing due in part to a combination of their prolonged longevity, loss of spouses and siblings in later life, and living away from or not having children (Suzuki, Dolley, and Kortt, 2021). Moreover, due to the rising number of unmarried or childless people in the country, in a death-laden, Japan, the size of the childless older population is also expected to grow at a rate unparalleled in the country's history (Park, 2020).

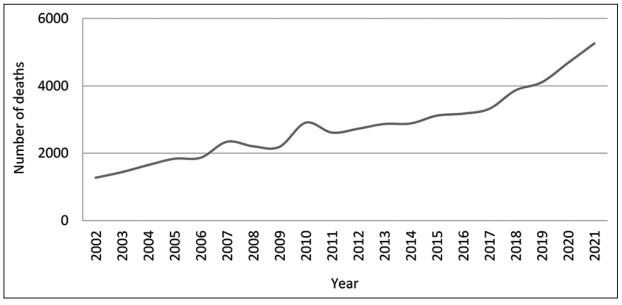

Figure 3. Number of those aged 65 or older who died at home alone and were investigated on the causes of deaths, Tokyo, Japan, 2002-2021.
Source: Tokyo Medical Examiner's Office (2022)

Under pressure from the coming of a death-laden society, as mentioned earlier, some policymakers in Japan may find the promotion of "dying in place" a necessary policy direction (Interview data, Tokyo, June 3, 2021). According to a hypothetical estimate for the case of Tokyo, if successfully promoted, a pursuit of "dying in place" could potentially mitigate the shortage in hospital beds by up to 30% by 2030 (Interview data, Tokyo, April 12, 2021). However, if rushing unprepared, this may, in turn, cause a surge in the prevalence of "lonely deaths," thus simply trading one major public health issue for another with which society must contend for decades to come. First, an increase in the incidence of "lonely deaths" directly threatens the comfort and dignity of those dying. In addition, it would result in increasing public health expenses for responding to a range of associated medical and legal needs. Hence, careful strategy and preparation are called for in promoting "dying in place." On the verge of becoming a death-laden society, Japan is immensely pressured to quickly develop an appropriate policy strategy to carefully and effectively rearrange the medical and social infrastructures for those with terminal health conditions to die in their preferred places in the way they wish.

3.4. Urgency to facilitate national discussions on end-of-life options

Japan must engage not only those in advanced age but also all citizens in a hard discussion on a subject that is highly controversial but may require swift policy consideration: End-of-life options. As discussed above, many of those in advanced age in the country desire "dying in place" in search of greater comfort and dignity during their last days. To prepare for the ballooning deaths in the coming decades, Japan today needs to solicit national consensus on whether and how the country may offer those dying a new end-of-life option in addition to what is available today.

Under Japan's current institutional framework, the only end-of-life option that is publicly acknowledged is to arrange "dignified death" (Spoden, 2020). One may seek "dignified death" by leaving advance directives for medical decisions before one's own death, which spells out his or her preferences regarding medical treatment in the event of terminal illness, falling into a state of coma, or reaching the late stages of dementia near the end of life (Japan Society for Dying with Dignity, 2022). Unlike euthanasia, this option does not involve any medical intervention to actively shorten – let alone actively terminate – the patient's life. With the option of "dignified death," the dying person may simply seek to die "naturally" by expressing their desire to refuse mere life-extending medical interventions. This way, the person may avoid unnecessary suffering and relieve caregivers of emotional and financial burdens caused by critical medical decision-making during moments of crisis or grief (Hirayama, Otani, and Matsushima, 2017). Over the past few decades, an increasing number of medical institutions and related organizations have come to consider "dignified death" as de facto legal. However, a lack of codified laws that explicitly justify this practice still leads many clinical doctors to remain wary of patients' requests to pursue this option (Interview data, Tokyo, 11 May 2021).

Meanwhile, other developed countries around the world have already moved forward to legalize end-of-life options that involve doctors' active role in responding to patients' desire to shorten their lives. One example is "physician-assisted suicide," a form of voluntary euthanasia that may be applied only to those suffering from terminal health conditions who consistently express a conscious desire to shorten their lives to end their sufferings (Kamm, 1999). Unlike euthanasia in its conventional form, this option involves doctors assisting a patient's suicide by providing the necessary means such as lethal doses of medication, or information about the means, for the patient to use to perform the life-ending act (Ahlzen, 2020). In recent years, an increasing number of countries around the world have legalized this option, including Switzerland, Belgium, the Netherlands, Luxembourg, Canada, Spain, and some regions of the US and Australia (Canetto and McIntosh, 2022). While a pursuit of individuals' rights to control their deaths is the proclaimed reason to legalize this end-of-life option, behind this trend, one of the root causes to call for access to this option stems from the steady aging of those countries' populations (Interview data, Tokyo, 22 February 2021).

To date, Japanese laws have never permitted any form of euthanasia including "physician-assisted suicide." Due in part to the lack of open public discussion about end-of-life options, neither the government nor medical professional communities have ever taken the initiative to formally discuss the possible importance of introducing this option, or anything similar, to Japan (Interview data, Nagoya, July 29, 2021). The lack of the discussion stems in part from a traditional cultural value held among the citizens, today's older generations in particular, that tends to regard death and dying as a highly private matter (Interview data, Osaka, February 14, 2021). The coming of a death-laden society will likely force the country to respond to the demands of an unprecedented number of older people potentially seeking to legalize "physician-assisted suicide" as a desirable option to be added to "dignified death." Despite this strong likelihood, however, the lack of national discussion to date indicates that Japan remains unprepared for facing this scenario.

3.5. Crematorium shortages and their culture impact

Against the backdrop of ballooning deaths in Japan, an unprecedented concern in the country's modern history has emerged regarding how individual bodies are handled after death: A growing shortage of crematoriums. While currently observed only in a limited number of urban areas, if persisting in the coming decades and growing nationwide in scale, crematorium shortages may also grow to become a broader cultural issue (Interview data, Tokyo, July 16, 2021). As the fifth area of challenge, Japan is pressured to address how the crematorium shortages may contribute to undermining a cultural legacy that has long supported remaining relatives in managing their grief and loss of the deceased.

The shortages have first caused a substantial delay in the timing of carrying out cremations in Tokyo and Osaka areas. Across Japan, since the early 20th century at least, a cremation has typically been held on the 3rd day after one's death (Suzuki, 2000). Over the past several years, however, a rising number of residents in those areas have come to wait for 10 days, even for 2 weeks in some cases, on a waiting list to cremate their deceased family. In the coming decade, such a delay will likely begin to occur in many other major cities across the country (Interview data, Tokyo, July 21, 2021).

This is a grim trend particularly to the case of Japan. While not legally mandated, cremation, as opposed to burial, has substantially been the only method available to clear away corpses since the early 20th century. Even today, in 2021, above 99.8% of all deceased bodies were cremated; by far the highest rate across the world, rendering Japan unique from an international perspective (Cremation Society, 2022). The shortages are also escalated by a steady decrease in the number of crematory facilities, particularly in urban areas, over the past decades. In 2000, there were about 2100 registered facilities across the country, but the number had dropped to nearly 1400 by year 2020 (Japan Association of Environmental Crematory, 2022). Behind this trend is active urban planning and renovations in which many cities have engaged particularly since the 1980s. Both city governments and funeral industries have experienced significant challenges in maintaining, let alone newly establishing, crematory facilities, and facing fierce opposition from local residents who are fearful of negative impacts that the presence of crematory facilities may create on the real estate value and public images of their communities (Interview data, Tokyo, November 8, 2020).

Crematorium shortages are more than a public health issue; this may contribute to eroding the national cultural framework that has long supported remaining relatives through the emotional challenges caused by their grief and loss of deceased family members: Family unity through funeral reunion. Conventionally, cremation is conducted in the late morning only an hour or two after the main funeral rite to make it easier for attendees from afar – immediate family members, remote relatives, friends, and coworkers – to attend the event and return home on the same day (Suzuki, 2000). In response to the emerging shortages, however, a growing number of families in Tokyo and Osaka areas are opting to cremate their deceased members outside the conventional hours while keeping the conventional date, 3 days after the death (Interview data, Osaka, 22 May 2020). Furthermore, some families choose to use funeral homes and morgues located in rural areas so as to carry out funerals in the conventional date and hours. Either way, despite their effort, an increasing number of those families have suffered from a lower turnout in the funeral attendees mainly because these alternative approaches interrupt some prospective attendees' daily schedules (Interview data, Tokyo, 12 March 2021).

A funeral provides many with an opportunity to hold a family reunion, which helps to give both the hosts and attendees a sense of intimacy and social bonding. This way, funerals have long served as a significant cultural event in which people emotionally support each other by sharing their grief, and thus mitigating their intense sense of loss (Tsuji, 2006). In the coming decades, the ongoing shortage in crematoriums will likely erode the role of such cultural legacies for an increasing number of families in urban areas, possibly across the country, by causing the need for funerals to be conducted outside the conventional time and space. While cultural impacts of this trend may be profound, to date, no specific policy measure has been discussed at the national level to address this prospective challenge (Interview data, Tokyo, March 18, 2021).

4. Conclusion: Call for the future research

Today, Japan is on the verge of another demographic transition that will significantly affect its society and culture – just in the coming decade, if not sooner, the world's first super-aged society will become the world's first death-laden society. This prospective transition deserves international attention as a new set of unique challenges for the death-laden society begins to manifest.

This paper has contributed to advancing the growing body of the literature on this emerging subject by exploring and outlining main areas of prospective policy challenges that Japan must swiftly address to mitigate the impact of this transition. The challenges outlined in this paper include but are certainly not limited to five areas of policy

concerns related primarily to the health, well-being, and comfort of citizens of all ages, but particularly those in advanced age and those living out their past days as well as their relatives. Together with the continuous shrinking of the working-age population, the ballooning of deaths – annual deaths of 1.6 million people – will likely exacerbate already existing shortages in clinical doctors and hospital beds and contribute to pushing to insolvency the national expenditures for medical and long-term care. These challenges will be inevitable especially if hospitals remain the primary place of Japanese citizens' death in the coming decades. Promoting "dying in place," however, will create the grim risk of a further prevalence of "lonely deaths" among those in advanced age and living in urban areas. Japan is also under pressure to begin swiftly increasing citizens' end-of-life options, such as physician-assisted suicide, should a majority of them call for it. Finally, but not of the least importance, a shortage of crematoriums needs to be effectively addressed. An immediate policy measure needs to be implemented not only for the sake of public health and sanitation but also to help preserve the cultural legacy that has long supported remaining family members in managing their grief and loss of the deceased.

Death-laden society is a newly emerging subject. To date, little systematic study has been done to discuss the impact of the coming of this new phase of demographic shift. Furthermore, this paper is based mainly on findings from exploratory research; thus, the discussions presented above are only limited and preliminary. More research, both empirical and theoretical, is called for to continue examining the way in which a death-laden society will affect the health, well-being, and comfort of those in advanced age in Japan, including and beyond the five areas of policy challenges discussed in this paper. Of particular importance for the future research is to pay close attention to the prospects of rural areas of the country, including remote islands, as a result of the transition into a death-laden society. These areas have experienced more rapid aging and population implosion than the rest of the country. Therefore, these areas will likely not only contend with greater challenges in the five policy areas as explored in this paper; the transition into a death-laden society may also generate challenges that are unique to these areas. As such, among the goals for the future research should be to uncover how unevenly the coming of a death-laden society affects those of advanced age and their relatives in large cities compared to rural and remote regions of the country.

Furthermore, important is to examine how the future changes to labor market institutions and the prospects for national economic vitality may be affected by Japan's transition into a death-laden society. The burst of the asset price bubble in the early 1990s weakened long-term employment security of workers of all ages, which has contributed to reduced productivity in some sectors of the labor force (Kamabayashi and Kato, 2016). Together with mounting pressure from the rapid aging of the population, Japan's fragile economic situation has led to austerity-motivated cuts to welfare policies and programs for older people, including public pensions and public health-care programs for the aged (Suzuki, Dolley, and Kortt, 2021). A thorough assessment of the impact of a death-laden society includes calls for close attention to the associated current and future prospects for Japan's national economic vitality and competitiveness within the ever more competitive global economy.

The future research should also draw policy suggestions to help mitigate the prospective risks and challenges of Japan's transition into a death-laden society. Such suggestions would also serve as a source for the current and future policymaking of other countries that may follow the path of a super-aged Japan to become death-laden societies in the conceivable future including South Korea, China, Taiwan, and Singapore. Specific and sound policy suggestions require future, more issue-specific research. Nonetheless, a possible direction that future policymakers might consider drawing from this paper is to expand the existing policy paradigm surrounding older people and later life. To date, the age-related policy goals in many countries commonly focus on protecting the health, well-being, and more broadly the quality of life of older people. The prospective challenges of a death-laden Japan, as explored in this paper, suggest that in the conceivable future, rapidly aging societies including Japan itself more explicitly include a goal of enhancing what may be referred to as the quality of dying. In a death-laden society, more and more people will likely call for societal support for protecting comfort and respecting dignity not only in the experience of the advanced stage of their lives but also during the very process of their dying. Such a policy direction should also aim to help younger people become less laden with various burdens brought on by the prospective ballooning of deaths in their society.

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RESEARCH ARTICLE

Unaccompanied refugee minors from the Democratic Republic of the Congo, Eritrea, Myanmar, and Somalia: Educational attainment, economic well-being, and social ties in the United States

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Abstract

In 2019, there were 21.3 million refugees around the globe. A small number of these are accepted to the United States each year under the Unaccompanied Refugee Minor Foster Care Program. There is currently limited research on the outcomes of young adults served through this unique program. In this paper, we share outcomes (educational attainment, economic well-being, and social ties) for young adults who leave care from the countries of the Democratic Republic of the Congo, Eritrea, Myanmar, and Somalia. The authors report descriptive statistics for young adults who discharged from the foster care program ($n = 388$) as well as Pearson's Chi-square tests to test correlations between outcomes and country of origin. Results show that youth from Myanmar is most likely to be enrolled in college at time of discharge. Youth from the DRC is equally likely to be enrolled in college or to have only completed a GED or high school diploma. Youth from Myanmar is more likely to be employed than youth from other countries. Eritrean youth was more likely to be lacking economic self-sufficiency at time of discharge than youth from other countries. Results from this study suggest ways that service providers can tailor service plans to help youth from different countries achieve the best outcomes, and pose questions for future research.

Keywords: Unaccompanied refugee minor; Foster care; Education; Social connections; Employment; Self-sufficiency

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1. Introduction

According to the 1951 Refugee Convention, refugees are a specific a group of people who have been forced to flee their homelands due to persecution (or the fear of persecution), based on their race, religion, political opinion, nationality, or membership in a particular social group (UNHCR, 2021).

The Unaccompanied Refugee Minor (URM) Foster Care Program was established in the 1970s. The URM program is a specialized foster care system that uses best practices

from the child welfare system and the refugee resettlement programs and accepts qualified immigrant and refugee children who lack parents or caregivers at time of entry to the U.S. The goal of the foster care program is to be culturally competent, offering cultural and religious community linkages to meet unaccompanied children's unique needs (United States Conference of Catholic Bishops [USCCB], 2013). To accomplish this, each URM program provides specialized training to foster parents and staff to help build a supportive environment for the children (USCCB, 2013). The URM program works to advance social justice through the view that "everyone deserves equal economic, political, and social rights and opportunities" (National Association of Social Workers [NASW], 2017, paragraph 2) and is designed to provide opportunities to refugee youth that is equal to those of U.S. born youth served through foster care (USCCB, 2013).

1.1. Study justification and literature review

The well-being of refugee children in their host countries is important to protect their human rights and advance their ability to participate in society. Unfortunately, there is little research on the outcomes of refugee children, especially those served specifically by the URM program. Luster *et al.* (2009) found that Sudanese youth served by the URM program struggled to adjust as foster parents wanted to have authority over the teenagers and they were used to living on their own in groups, and that misunderstandings around cultural traditions only created deeper conflict and struggles in communication. A study by Socha *et al.* (2016) found that Eritrean youth had vastly different expectations before arrival as compared to what they found when they entered the foster care program, leading to discontent with the program initially and distrust in their social networks that had fueled initial expectations of resettlement. Evans *et al.* (2021) looked at youth (both immigrant and refugee) served by the US URM program to assess factors that influence self-sufficiency on discharge. They found that employment, English proficiency, and educational attainment all had significant relationships with self-sufficiency (Evans *et al.*, 2021). Due to the dearth of literature out of the United States, at times, researchers and service providers rely on research about the integration of refugee children in other countries such as Sweden (Wimelius *et al.*, 2017), Canada (Baffoe, 2011), Ireland (Horgan & Ni Raghallaigh, 2019), and Britain and Europe (Bell, 2005).

When looking at youth served by the URM program at large, there is also research into the outcomes that focuses specifically on youth from Central America. For example, youth's educational outcomes have been assessed for youth from Honduras, Guatemala, and El Salvador (Crea *et al.*, 2017), and employment outcomes have been assessed for youth from Honduras, Guatemala, and El Salvador (Hasson 3rd *et al.*, 2021). However, the experiences in home country, migration journey, benefits available on arrival to the US, and discriminations faced in the US are vastly different for youth who enters as refugees as compared to those who enter as undocumented immigrants through the South US border. Therefore, this study is looking at education, employment, and social ties (dimensions known to be important in terms of immigrant integration) for refugee youth from Myanmar, the Democratic Republic of the Congo, Eritrea, and Somalia as there are little quantitative data on these youth and they are some of the other refugee countries that are well represented in the dataset. The literature noted above that compares youth from different Central American countries has been utilized by social service providers in helping to tailor case plans and make inferences about what services may be more or less beneficial to different youth. Therefore, we propose a similar methodology and compare outcomes across the largest groups of refugees in the URM program.

According to Calvo *et al.* (2016), integration consists of three interrelated dimensions: Social, economic, and political. Therefore, this study aims to examine the indicators of social and economic participation, as there was no measure of political involvement in the administrative dataset. Others authors support the value of social connections in the lives of immigrants and show that it can lead to better mental health and well-being (Ager & Strang, 2008; Elsayed *et al.*, 2019; Revens *et al.*, 2021; Puyat, 2013) and ease the transition to a new environment (Bankston CL 3rd, 2014). We suggest that education is one aspect of social participation in line with Calvo *et al.* theory above. The literature widely supports the importance of education in terms of immigrant integration (Ager & Strang, 2008; Coomans, 2018; Naidoo, 2009). Similarly, a wide body of research supports the value that employment plays in integration and community involvement for refugees (Ager & Strang, 2008; Capps *et al.*, 2015; Hasson 3rd *et al.*, 2021; Mirembe *et al.*, 2019; National Academies of Sciences *et al.*, 2015). Self-sufficiency is a common metric used by the Office of Resettlement to measure the success of refugees in the US (Halpern, 2008). Self-sufficiency goes beyond holding a job and Halpern (2008) noted that some of the challenges to self-sufficiency include lack of resources, transportation, and language barriers.

1.2. Research questions

This study is guided by three research questions, focusing on the differences in county of origin for youth served through the URM program. They are as follows: RQ1: What are the levels of educational attainment for youth from the

Democratic Republic of the Congo, Eritrea, Myanmar, and Somalia? RQ2: What are the employment outcomes for youth from the Democratic Republic of the Congo (DRC), Eritrea, Myanmar, and Somalia? and RQ3: What social ties exist for youth from the Democratic Republic of the Congo, Eritrea, Myanmar, and Somalia?

2. Data and methods

2.1. Data source

Lutheran Immigration and Refugee Service (LIRS), one of the national agencies that administer the URM program, collects administrative data on each youth at the time that they discharge from the URM program. The administrative data is cross-sectional and contains a series of youth outcomes such as living situation, employment status, educational attainment level, English language skills, access to health care, and more. This information has been shared with the university and secondary data analysis was approved through the University of Maryland Baltimore County (UMBC's) Institutional Review Board. The sample for this study includes 392 youth who was discharged from an LIRS URM program during Federal Fiscal Year (FFY) 2015 or FFY2016.

The majority, 72.70% (n = 285), of the youth who exited URM care were male as compared with 27.30% (n = 107) females. Of the 392 youth in the sample, 25.77% (n = 101) are from Honduras, 19.90% (n = 78) are from Guatemala, 10.20% (n = 40) are from Mexico, 8.67% (n = 43) are from the Democratic Republic of Congo, 5.87% (n = 23) are from Myanmar, 5.36% (n = 21) are from each El Salvador and Somalia, 4.08% (n = 16) are from Eritrea, 1.53% (n = 6) are from Sudan, 1.28% (n = 5) are from Nepal, and 1% or less (n = 4 or less) are from the each of the following countries: Afghanistan, Belize, Bhutan, Cambodia, China, Ecuador, Ethiopia, Ghana, Haiti, India, Iran, Iraq, Ivory Coast, Kenya, Liberia, Nicaragua, Nigeria, Pakistan, Rwanda, Tanzania, and Thailand. The present study focuses on youth from DRC, Myanmar, Somalia, and Myanmar as they are the four countries with the highest number of refugees in the sample. The other countries with high numbers of youth (Honduras *et al.*) do not have refugee status, and many of these outcomes have been studied (Crea *et al.*, 2017; Hasson 3rd *et al.*, 2021). The average length of stay in the URM program for the youth in the study was 1002.77 days, or a little under 3 years.

Within the sample, the majority of URM youth discharge from the foster care program during the young adulthood years of 18 – 23. The age of majority for foster care services varies by state, and URM programs are located across the United States and at the time of data collection were in Arizona, California, Colorado, Washington DC, Massachusetts, Michigan, Mississippi, New York, North Dakota, Pennsylvania, Texas, Utah, Virginia, and Washington state. Only 1.78% (n = 7) of youths who discharged from the URM program were below the age of 14 years. Five youths (1.28%) were 15 years of age, 0.77% (n = 3) of youths were 16, and 2.04% (n = 8) of youths were 17 years old. The majority were aged 18 or over at the time of discharge: 12.75% (n = 50) were 18, 22.19% (n = 87) were 19, 16.07% (n = 63) were 20 years old, 32.40% (n = 127) were 21, 4.34% (n = 17) were 22, 3.06% (n = 12) were 23, and 0.51% (n = 2) were 24 years of age. For 2.30% (n = 9) of youths, the date of birth was missing and so age at discharge could not be calculated. The average age at discharge was 19.7 years old.

2.2. Data management

The researcher assessed the dataset for missing data and worked with staff at LIRS to fill in as many fields as possible. Any data that appear inconsistent were questioned and rectified to ensure data integrity of the sample. A codebook was created for the dataset which lists all possible responses and assigned a numeric value for nominal variables, for ease of data analysis. Finally, all string variables were transformed into numeric values and the data were imported to Stata 14SE for analysis. List-wise deletion was used to eliminate incomplete files as there were very few; the final sample was 388 youths.

2.3. Measures of analysis

The outcome variables of interest were level of education, economic well-being, and social ties. The youth's highest level of education was recorded as one of the following: (1) K-12; (2) GED, high school diploma, attending a vocational technology program, or attending an associate's degree program; and (3) attending a 4-year college.

Two different measures of economic well-being were considered in this analysis: Employment status and self-sufficiency. Employment status was recorded for each URM youth at the time of discharge and for the purposes of this analyses was dichotomously coded as (1) unemployed if the response was: Unemployed, no work authorization, not employed due to disability, or attending school full time and unable to work; or (2) employed if the response was employed part time or employed full time. Self-sufficiency was dichotomously coded as (1) not self-sufficient if the response was no income or income limits standards of living or (2) self-sufficient if the response was income meets basic needs, self-sufficient, or income is beyond enough.

Social ties were measured by the youth's connections to supportive adults. The literature strongly suggests that

positive adult roles and mentors are beneficial to both youths in foster care (Avery, 2011), immigrant youth (Rossiter & Rossiter, 2009), and most specifically immigrant youth in foster care (Evans *et al.*, 2022; Socha *et al.*, 2022). For youth who exited care in FFY2015, the variable was yes or no, where the caseworker determined if the youth had a connection to a positive adult role model. For youth who discharged in FFY2016, the options were more inclusive, using a Likert scale to rate both the quality and quantity of these adult relationships. Therefore, the responses for youth who existed care in FFY2016 were simplified to a yes/no response for analysis. To do so, any youth who scored a 1 "youth has no social connections" for either the quantity or quality measure were coded as (2) no; and for youth with a score of 2 or more for both quality and quantity of social connections they were coded as (1) yes. In addition, the variable that describes the youth's living situation on discharge is being used to describe social connections. This was dichotomously coded as (1) living with others if the response was living with friends, relatives, former foster family, adoptive family, in college, or in a residential program or (2) living alone if the response was living alone, or homeless. Youths who were living in a residential treatment center or incarcerated were eliminated from this analysis.

2.4. Analysis methods

Descriptive statistics were utilized to summarize the characteristics of the sample. Then, the author used cross-tabulations and Pearson's Chi-square tests through Stata 14SE to assess the significance of relationships between variables. The relationship of each country: Myanmar, Democratic Republic of the Congo, Eritrea, and Somalia, as compared with all "other" countries was individually assessed in comparison to the youth's status at discharge for the level of education, employment status, self-sufficiency, social connections, and living status at discharge.

3. Results

3.1. Educational attainment outcomes

The educational outcomes for youth served by the URM program vary greatly. At discharge, 56.52% (n = 221) of all youths were still enrolled in a K-12 setting, 24.81% (n = 97) had completed high school, a GED, or were in a vocational technology or associate's degree program, and only 18.67% (n = 73) were enrolled in a bachelor's degree program. For the level of education, there is a statistically significant relationship with youth from Myanmar X^2 (2, 388) = 24.09, $P < 0.001$ when compared to youth from other countries. As shown in Table 1, of the youth from Myanmar, they are most likely to be enrolled in college. Similarly, for youth from the Democratic Republic of the Congo when compared to youth from other countries, there was a significant relationship with level of education χ^2 (2,388) = 14.45, $P < 0.01$. Youths from the DRC are equally likely to be enrolled in a college or have a GED/HS diploma or be attending a certificate program at the time of discharge from foster care. There were no significant relationships found between educational outcomes and youth from Somalia or Eritrea.

The analysis was also run with all countries of interest in one model, as displayed in Table 2. This shows an overall statistically significant relationship between country of origin and educational outcomes, χ^2 (8, 388) = 39.34, $P < 0.001$. Youths from Myanmar are most likely to be enrolled in college at discharge. Youths from Somalia, Eritrea, and other countries are most likely to still be enrolled in a K-12 education. Congolese youths are fairly evenly distributed across all three educational outcomes.

3.2. Economic well-being outcomes

More than half, 60.99% (n = 233) of the youths in the sample were employed and 62.23% (n = 117) were self-sufficient. There is a statistically significant relationship between Burmese youth (those from Myanmar) and employment X^2 (1,379) = 4.37, $P < 0.05$, showing that they are more likely to be employed (Table 3). There were no statistically significant differences between expected and actual counts found between employment and youth from the DRC, Eritrea, or Somalia.

However, the analysis for employment was also run with all of the countries of interest in one model, and this did show a statistically significant relationship of χ^2 (4,379) = 10.09, $P < 0.05$ as displayed in Table 4. In this analysis, youth from other countries and those from Somalia were about equally likely to be employed versus not employed, whereas youths from Myanmar and DRC were more likely to be employed and youths from Eritrea were less likely to be employed.

A statistical model was run to examine each country of origin as compared with other countries. The only significant finding at the time was that of Eritrean youth, however, the cell sizes are too small to be valid. Therefore, the results show no significant differences between expected and actual counts found between self-sufficiency and youth from Myanmar, DRC, and Somalia. However, when all countries of origin were compared in one analysis (Table 5), there was a statistically significant relationship between country of origin and self-sufficiency, X^2 (4,188) = 13.76, $P < 0.01$. Burmese, Congolese, and Somali youths were more likely to be self-sufficient as compared to not self-sufficient. Youths from other

Table 1. Educational outcomes by each country of origin (*n* = 388).

Country of Origin	K-12	GED or HS Diploma or 2 years	4 years college	Pearson Chi-squared
Analysis 1				24.09***
Myanmar	2 (8.7%)	10 (43.48%)	11 (47.83%)	
Other	216 (59.18%)	87 (23.84%)	62 (16.99%)	
Analysis 2				11.71**
DRC	10 (29.41%)	12 (35.29%)	12 (35.29%)	
Other	208 (58.76%)	85 (24.01%)	61 (17.23%)	
Analysis 3				1.08
Eritrea	11 (68.75%)	3 (18.75%)	2 (12.50%)	
Other	207 (55.65%)	94 (25.27%)	71 (19.09%)	
Analysis 4				0.36
Somalia	12 (57.14%)	6 (28.57%)	3 (14.29%)	
Other	206 (56.13%)	91 (24.80%)	70 (19.07%)	

*P<0.05; **P<0.01; ***P<0.001

Table 2. Educational outcomes across countries of origin (*n* = 388).

Country of Origin	K-12	GED or HS diploma or 2 years	4 years college	Pearson's Chi-squared
Myanmar	2 (8.7%)	10 (43.48%)	11 (47.83%)	
DRC	10 (29.41%)	12 (35.48%)	12 (35.29%)	
Eritrea	11 (68.75%)	3 (18.75%)	2 (12.50%)	
Somalia	12 (57.14%)	6 (28.57%)	3 (14.29%)	
Other	183 (62.24%)	66 (22.45%)	45 (15.31%)	
				39.34***

*P<0.05; **P<0.01; ***P<0.001

countries were evenly distributed and Eritrean youths were more likely to be not self-sufficient.

3.3. Social ties outcomes

Almost all, 96.90% of youth (n = 375) were determined to have sufficient social connections with positive adult role models and 90.19% of youths were living with others. For the subsample of youth for whom more detailed data were available, 79.79% (n = 154) of youths were said to have a sufficient quantity of social connections and 20.21% (n = 39) did not. About 87.89% (n = 167) of youths in the subsample from FFY2016 had sufficient quality in their social connections. There was no statistically significant relationship for youth of any country when looking at social connections. When looking at the youth's living situation on discharge as a measure of their social connectedness, there were no statistically significant relationships for youth from any country.

Table 3. Employment outcomes by each country of origin.

Country of Origin	Not employed	Employed	Pearson's Chi-squared
Analysis 1			4.37*
Myanmar	4 (18.18%)	18 (81.82%)	
Other	145 (40.62%)	212 (59.38%)	
Analysis 2			2.20
DRC	9 (27.27%)	24 (72.73%)	
Other	140 (40.46%)	206 (59.54%)	
Analysis 3			3.76
Eritrea	10 (62.50%)	6 (37.50%)	
Other	139 (38.29%)	224 (61.71%)	
Analysis 4			0.12
Somalia	9 (42.86%)	12 (57.14%)	
Other	140 (39.11%)	218 (60.89%)	

*P<0.05; **P<0.01; ***P<0.001

Table 4. Employment outcomes across countries of origin (*n* = 379).

Country of Origin	Not employed	Employed	Pearson's Chi-squared
Myanmar	4 (2.68%)	18 (7.83%)	
DRC	9 (6.04%)	24 (10.43%)	
Eritrea	10 (6.71%)	6 (2.61%)	
Somalia	9 (6.04%)	12 (5.22%)	
Other	117 (78.52%)	170 (73.91%)	
Total	149 (100%)	230 (100%)	X^2 = 10.09*

*P<0.05; **P<0.01; ***P<0.001

Table 5. Self-sufficiency outcomes across countries of origin (*n* = 188).

Country of Origin	Not self-sufficient	Self-sufficient	Pearson's Chi-squared
Myanmar	0	6 (5.13%)	
DRC	5 (7.04%)	13 (11.11%)	
Eritrea	9 (12.68%)	2 (1.71%)	
Somalia	4 (5.63%)	9 (7.69%)	
Other	53 (74.65%)	87 (74.36%)	
			13.76**

*P<0.05; **P<0.01; ***P<0.001

4. Discussion

Calvo *et al.* (2016) stated that the three interrelated dimensions of immigrant integration are social, economic, and political. Ager and Strang (2008) dig a little deeper and note that the markers of immigrant integration are employment, housing, education, and health, while facilitators of integration

include social connection, language and cultural knowledge, and safety. In this paper, we explore outcomes that both theories use to understand how unaccompanied minors from the DRC, Myanmar, Somalia, and Eritrea are doing after discharge from foster care in terms of educational attainment, employment status/self-sufficiency, and social ties. The results discuss differences among youth from different countries to help inform casework practices and the knowledge base in terms of how this unique group of youth is faring in the US after resettlement. Understanding well-being of vulnerable youths who are both immigrants and living in foster care is important to study to help achieve equity and well-being in society.

This analysis found a significant relationship between country of origin and education showing that youths from Myanmar are most likely to be enrolled in college at discharge whereas youths from Somalia, Eritrea, and other countries are most likely to still be enrolled in a K-12 education. The educational resilience, perseverance, and aspirations of refugee youth have been found to serve as a protective factor in the process of adjustment to a new country (Kumi-Yeboah & Smith, 2016; Kohli, 2011).

There was a significant relationship found between country of origin and employment. More specifically, youths from Myanmar and DRC were more likely to be employed; and youths from Eritrea were less likely to be employed at the time of discharge from the URM foster care program. At times, people need to move to find adequate employment; however, the dataset used for this study is cross-sectional at time of discharge and does not account for any internal migration that may occur in the months after leaving the foster care program. Mirembe *et al.* (2019) found that young adults in Uganda often moved to the cities to seek employment, and those aged 18 – 22, females, and those without children were more likely to be self-employed. Our study did not look at self-employment outcomes, but this could be an interesting area for future research.

Because self-sufficiency is a common metric used to assess success of refugees in the US (Halpern, 2008), it is included here as an indicator of economic well-being. Burmese, Congolese, and Somali youths were more likely to be self-sufficient as compared to not self-sufficient. When looking at the outcomes for employment and self-sufficiency, they seem to mirror each other in that those who are employed are also likely to be self-sufficient, perhaps due to the income from their employment. Evans *et al.* (2021) also found that employment and self-sufficiency were closed related outcomes.

There were no statistically significant relationships among any of the variables assessed for youth from Somalia. Somali refugees have been arriving to the US for many years and over 82,000 arrived between 1983 and 2007 (Betancourt *et al.*, 2015). Therefore, the Somali youth may have access to a more established cultural network, and therefore, greater supports available to them on arrival to the US than refugees from some of the other countries analyzed, which are newer populations. Social networks provide many benefits to immigrants including better mental health outcomes (Elsayed *et al.*, 2019; Revens *et al.*, 2021) and lower risk of food insecurity (Freiria *et al.*, 2021).

4.1. Limitations

The main limitation to this study is that the variables are from administrative data at an agency and standardized measures were not used to collect data on social connections or self-sufficiency. Standardized measures would be more efficient than relying on a caseworker to use a single item to assess these constructs. In addition, Chi-square analyses are built on the assumption that there is greater than 5 cases in each cell, and for some of the analyses above, this was not the case due to limited number of youth from these countries. Due to the small sample sizes from each country of origin, more in-depth statistical analyses of these data are not appropriate at this time. We do suggest that the social service agencies continue to track data and continue to coproduce statistical analyses in future years so that more than Chi-square analyses can be completed. For the time being, readers should use caution when interpreting the results of this study and use the knowledge to question what they see in the field and inform themselves rather than to make blanket assumptions about URMs from different countries.

4.2. Implications

Overall, the findings from this study help to support the idea that children from different countries have different experiences and, therefore, may have different outcomes. Service providers can use this knowledge when working with youth from these countries to better predict what help youth from a certain country may need, in relation to youth from other countries. Service plans for youth in foster care are typically individualized and this can help agency staff to predict what needs will be based on the ethnicities enrolled in their program. For example, knowing that Eritrean youths have different educational and employment outcomes can help in service planning before emancipation. This could include additional life skills programming for these youth, more targeted support in case planning 1 year from emancipation or targeted support in obtaining a job before emancipation.

This preliminary study shows that more research is needed. First, future analyses should include analysis of more demographics such as gender, length of stay in foster care,

and English level to determine if they have an influence on these outcomes as well as consider confounding outcomes such as mental health that can influence well-being in these areas. Importantly, qualitative studies could take place to ask questions and explain some of the findings in this paper such as why educational attainment is lower for Somali and Eritrean youth as compared to others. Understanding the root causes of these struggles could help URM programs to seek funding and build interventions that will assist a larger number of youth in reaching successful outcomes. Second, by pooling data over the years, or working collaboratively with the other federally granted agency that provides URM care, we could possibly increase the number of youth and, therefore, conduct similar analyses for youth of other countries of origin. If we had larger sample sizes through these methods, regression analyses and structural equation modeling could be used to better assess the relationship between these variables as it is possible that multiple of them could have influences in various ways.

5. Conclusion

This study provides descriptive statistics and correlations to help us understand some basic outcomes for youth from Eritrea, Somalia, the DRC, and Myanmar, which are countries with some of the highest enrollment rates in the URM program. Results show that youths from Myanmar are most likely to be enrolled in college and employed at time of discharge when compared to youth from other countries. Youths from the DRC are equally likely to be enrolled in college or to have only completed a GED or high school diploma. Eritrean youths were more likely to be lacking economic self-sufficiency at time of discharge than youth from other countries. The study found no statistically significant findings when it comes to social ties. While these findings are interesting and can assist case managers in adapting service plans for youth of specific backgrounds, they also provide questions for future research.

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Conflict of interest

The authors have no conflicts of interest to declare.

Author contributions

Conceptualization: Kerri Evans
Formal analysis: Kerri Evans
Writing – original draft: Kerri Evans
Writing – review & editing: Hannah Ferguson
The data owned by Hannah Ferguson.

Ethics approval and consent to participate

University of Maryland Baltimore County IRB approved secondary data analysis for this study.

Consent for publication

Hannah Ferguson, staff member of LIRS and owner of the administrative data set provided consent for publication.

Availability of data

As this data is administrative in nature and stems from files for youth in foster care, this data is not publicly available.

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ACCSCIENCE PUBLISHING

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RESEARCH ARTICLE

Used (Lived) versus Offered (Plain) densities of human settlement in space: An instance of the probabilistic consumption model

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Abstract

To people living in areas, the denser is the area, the more numerous are the opportunities of interpersonal and social interaction, of employment and of amenities of all kinds. The spatial density of human settlement is basically studied according to places, that is, area weighted. The notion of population-weighted density, or lived density, puts human density in the perspective of the people that experience it. Considering, respectively, the land units and the people as statistical populations of their own, the article provides a probabilistic model of human density in a geographical space, as a random variable in each statistical population, with specific probability density functions (PDFs) and cumulative distribution functions. The PDF of lived, "Used density" is derived from that of the plain, "Offered density" through a consumption model: Thus, their relationship is a specific instance of a well-established probabilistic model. The average used density is systematically larger than its offered counterpart: The ratio amounts to one plus the squared coefficient of variation of offered density. The relation between the two statistical distributions is illustrated using a Lorenz curve; the associated Gini index constitutes an indicator of population heterogeneity in a geographical space. A case of France's population as of 2019 is studied to demonstrate the methodology.

Keywords: Spatial heterogeneity; Density metrics; Land units; Consumption model; Lorenz curve; Gini index

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1. Introduction

The spatial density of human settlements stands out as a prominent feature of territories. Urban areas are endowed with high density of population and jobs: Typical urban residential densities range from 1000 inhabitants per square km to more than 100,000 in the densest parts of some Asian megacities (e.g., Dhaka). Rural areas, in contrast, exhibit sparse human settlements and low densities: Typical values of rural density range from near zero to some hundred inhabitants per square km (Aliaga *et al.*, 2015; Vorobyev, 2019). Thus, density is a relevant indicator of urbanization, together with the overall area population which is the primary indicator of city size in human geography. Spatial maps exhibiting zones colored according to their respective levels of density make a basic tool to understand the spatial structure of territories (e.g., Dijkstra & Poelman, 2014).

Yet, the basic indicator of human density pertains, essentially to space. As geographical space is likely to exhibit heterogeneity of human occupation, the average plain density (rather than "crude density" as worded by Craig, 1984) does not indicate much about the effective intensity of human occupation that people experience in their everyday life, in the places that they frequent – home, workplace, etc. This is why alternative, population-weighted indicators of density have been developed by pioneering researchers in the mid-1970s. Stairs (1977a; 1977b) introduced the person-average density as "the local density experienced by people, that is, spatial density of human population weighted by the number of people experiencing it." This indicator was called population-weighted (arithmetic mean) density by Craig (1984). The name of "population-weighted density" has popularized among geographers and economists that have used the indicator to understand "density as lived by people" and its effects on urbanization (Eidlin, 2010, Florida, 2012), mobility practices (Barnes, 2001), as well as greenhouse gas (GHG) emissions of travel (Lee & Lee, 2014). As stated by Barnes (2001: p.16), "we are interested in human behavior; what we want to know is what people perceive density to be: This would be more closely captured by giving equal weight to each person, rather than to each square mile of land…. thus (we shall) use a new measure called 'perceived density,' which is defined as a weighted average of traffic zone densities, where each zone is weighted by the number of residents." Ottensmann (2018a) provided a comprehensive literature review of the concept, its applications and the related issues, together with a diachronic application to major US cities.

So far, the concept of "lived density" and "population-weighted density" has been expressed using mathematical formulas of weighted averages involving the numbers of people living in pre-defined zones (Barnes, 2001; Craig, 1984; Ottensmann, 2018a; Stairs, 1977). The relationship between lived and plain density averages has been established by Lewontin & Levins (1989) and Ottensmann (2018a) using lengthy proofs. This article is aimed to state plain density and lived density in the standard probabilistic framework, involving statistical populations, respectively, of land units and people, local human density as a random variable in either statistical population, their respective probability density functions (PDFs), and the general relation between them. This relation consists in a specific instance of a well-known probabilistic model: The "consumption model" that arises in various fields from economics to traffic theory. To emphasize the consumption model that relates plain density to lived density, we consider land units as servers of spatial settlement for people. Then, plain density is also the offered density, whereas lived density is also the "used density," since people are the users of the settlement services. The relation between the PDFs enables one to build average indicators in a straightforward way. It also allows for considering classical indicators of heterogeneity in a simple way: we introduce interquartile ratios and above all the Lorenz curve and the Gini index of human density in a given geographical space.

The rest of the article is organized in four parts: After stating the methodology, we will apply it to a case study of communal density in France as of 2019, before providing a discussion and a short conclusion. Four appendices provide more details on (A) the notation table, (B) the consumption model, (C) the lognormal distribution and its use for consumption models, and (D) the linear-log model of used density CDF.

2. Theoretical background

We shall first define land units to deal with a geographical space as a statistical population of such elementary places (§2.1). Then, human density is put as a random variable with its own probabilistic distribution, mean and other statistical moments, and heterogeneity measures (§2.2). The next step is to shift the statistical perspective from the statistical population of land units to that of people: The random variable of used human density inherits its probabilistic features – probability distribution and statistical moments including the average value – from its offered counterpart on the basis of a consumption model (§2.3). We then recall the Lorenz curve and the Gini index as classical tools for inequality measurement and adapt them to the inequality of human population among land units (§2.4). Finally, we recall some previously proposed indicators of heterogeneity for human density and we restate them using our notation (§2.5).

2.1. Geographical space as a statistical distribution of land units

2.1.1. Territory, zoning system, and population

To analyze human density in a geographical space, it is convenient to model that space using a set Z of zones z. Each zone has its own ground area, A_z, and human population, P_z. Its spatial density of human population is simply:

$$x_z = P_z / A_z \quad (1)$$

The territory has total ground area of $A_Z = \sum_{z \in Z} A_z$ and total population of $P_Z = \sum_{z \in Z} P_z$. Its human density averaged over space is therefore

$$\bar{x}_Z = P_Z / A_Z \quad (2)$$

2.1.2. Spatial units as a statistical population

When zones are used to analyze the statistical distribution of some spatial variables in a discrete way, they are often called "spatial units." Here, we shall rather refer to zones as "spatial entities" and define "land units" as elementary places of unit ground area, say a_1. Such land units are more convenientthan zones to constitute the statistical population of places since, being identical in area, it is easier to compare them in other respects such as the human population.

The assignment of land units o to any zone z is an idealization: Thinking of the unit ground area a_1 as 1 square km or 1 hectare, we expect most zones to involve a non-integer number of land units. We nevertheless denote as " $o \in z$" the composition of zone z out of land units o. To every land unit o, with population denoted by p_o, is associated a human density as follows:

$$x_o = \mathrm{p}_o / \mathrm{a}_1 \tag{3}$$

Notionally, the zone area adds up those of the land units in it, and similarly, the zone population adds up those of its land units:

$$\mathrm{A}_z = \sum_{o \in z} \mathrm{a}_1 \tag{4a}$$

$$\mathrm{P}_z = \sum_{o \in z} \mathrm{p}_o \tag{4b}$$

2.2. Human density in the statistical population of land units

Human density x constitutes a random variable in the statistical population of land units, with PDF and CDF denoted by f_O and F_O, respectively.

2.2.1. Average human density over space

The average human density over space stems from the probability density function f_O in the usual way (e.g., Blitzstein & Hwang, 2015):

$$\bar{x}_{\mathrm{O}} \equiv \int x.\mathrm{f}_{\mathrm{O}}(x)dx \tag{5}$$

This general version of average human density over space is equivalent to the discretized one: Denoting as $\bar{\mathrm{O}}$ the total number of land units, it holds that:

$$\bar{x}_{\mathrm{O}} \equiv \frac{\sum_{o \in \mathrm{O}} x_o}{\bar{\mathrm{O}}}$$

As $\bar{\mathrm{O}}.\mathrm{a}_1 = \mathrm{A}_Z$, replacing x_o with p_o/a_1 and $\mathrm{a}_1\bar{\mathrm{O}}$ with A_Z due to (4a) aggregated over zone set Z, it comes out that

$$\bar{x}_{\mathrm{O}} = \frac{\mathrm{P}_Z}{\mathrm{A}_Z} \tag{6}$$

Thus $\bar{x}_{\mathrm{O}} = \bar{x}_{\mathrm{Z}}$, as could be expected.

2.2.2. On the statistical moments of human density

Higher order moments of human density in the statistical population of land units are defined in the usual way (e.g., Blitzstein & Hwang, 2015): At order r,

$$\mathrm{E}_{\mathrm{O}}[x^r] \equiv \int x^r.\mathrm{f}_{\mathrm{O}}(x)dx \tag{7}$$

Under the idealized assumption that human density would be homogenous among the land units composing any zone (i.e., no intra-zonal heterogeneity of density), then $\mathrm{E}_{\mathrm{O}}\left[x^r\right] = \mathrm{E}_{\mathrm{Z}}[x^r]$, wherein the inter-zone average E_Z of density moment x^r is defined as

$$\mathrm{E}_{\mathrm{Z}}[x^r] \equiv \sum_{z \in \mathrm{Z}} \frac{\mathrm{A}_z}{\mathrm{A}_{\mathrm{Z}}} \left(\frac{\mathrm{P}_z}{\mathrm{A}_z}\right)^r \tag{8}$$

The formula enables one to calculate $\mathrm{E}_{\mathrm{O}}\left[x^r\right]$ in an exact way under intra-zonal homogeneity of density, or in an approximate way otherwise.

2.2.3. Indicators of offered human density heterogeneity

Local human density is likely to be heterogeneous among land units, even inside every zone z. The intra-zone variance of human density is a metric for that heterogeneity within z. It is defined as $\mathrm{V}_{\mathrm{O}}^{(z)}\left[x\right] \equiv \mathrm{E}_{\mathrm{O}}\left[x^2 | o \in z\right] - (\mathrm{E}_{\mathrm{O}}\left[x | o \in z\right])^2$, and satisfies that

$$\mathrm{V}_{\mathrm{O}}^{(z)}\left[x\right] = \left\{\sum_{o \in z} \frac{\mathrm{a}_1}{\mathrm{A}_z} x_o^2\right\} - x_z^2 \tag{9}$$

Over the territory, the overall variance of human density can be measured using the law of total variance, that is, its decomposition into within-group variance and between-group variance (e.g., Blitzstein & Hwang, 2015):

$$\mathrm{V}_{\mathrm{O}}\left[x\right] = \left\{\sum_{z \in \mathrm{Z}} \frac{\mathrm{A}_z}{\mathrm{A}_{\mathrm{Z}}} \mathrm{V}_{\mathrm{O}}^{(z)}\left[x\right]\right\} + \left\{\sum_{z \in \mathrm{Z}} \frac{\mathrm{A}_z}{\mathrm{A}_{\mathrm{Z}}} (\bar{x}_z - \bar{x}_{\mathrm{O}})^2\right\} \tag{10}$$

The associated standard deviation (SD) and relative dispersion (coefficient of variation, CV) are therefore

$$\mathrm{SD}_{\mathrm{O}}\left[x\right] \equiv \sqrt{\mathrm{V}_{\mathrm{O}}\left[x\right]} \tag{11a}$$

$$\gamma_{\mathrm{O}}\left[x\right] \equiv \mathrm{SD}_{\mathrm{O}}\left[x\right] / \bar{x}_{\mathrm{O}} \tag{11b}$$

In empirical distributions, the variance and, in turn, the SD and CV are sensitive to outliers, that is, values falling out of the ordinary range of the variable. As the quantile values $\mathrm{F}_{\mathrm{O}}^{(-1)}(\alpha)$ at probability level α neither too small nor

too high are less sensitive to extreme values, they are taken as "robust statistics" (Wonnacott & Wonnacott, 1990). Thus, for non-negative real variables, the ratios between corresponding pairs $F_O^{(-1)}(\alpha)$, $F_O^{(-1)}(1-\alpha)$ constitute robust indicators of heterogeneity in the distribution: These include the inter-quartile ratio at level $\alpha = 1/4$ as well as the inter-decile ratio at level $\alpha = 1/10$.

For instance, a log-normal distribution LN (m,s^2) yields the following interquartile ratio at order α (cf. appendix C), with $\Phi^{(-1)}$ the inverse CDF of the reduced Gaussian variable:

$$\frac{F_O^{(-1)}(1-\alpha)}{F_O^{(-1)}(\alpha)} = e^{2s\Phi^{(-1)}(1-\alpha)}$$

2.3. Human density as lived by the statistical population of inhabitants

Any individual u inhabits a zone z_u and inside it a land unit $o(u)$. One defines the "used density" or "lived density" as the human density in the land unit inhabited by the user:

$$x_u \equiv x_{o(u)} \tag{12}$$

Among the population of users, of size $\bar{U} = P_Z$, the user-centric density is a random variable, with specific PDF and CDF denoted, respectively, as f_U and F_U.

2.3.1. Relationship between f_o and f_U

The PDF f_U of used human density is related to that f_O of offered human density by a "consumption model" well-known in probabilistic theory, especially in renewal theory (Kleinrock, 1975):

$$f_U(x) \propto x.f_O(x) \tag{13}$$

The reason is that the land units of which the human density belongs to $[x, x+\delta x]$, in proportion $f_O(x).\delta x$ in their distribution, do contain $x.a_1$ users each: Hence, their total number of users amounts to $a_1.x.f_O(x).\delta x.\bar{O}$. These users are those with user-centric density in $[x, x+\delta x]$ and those users only: Thus, their number is also $f_U(x).\delta x.\bar{U}$. On combining both formulas, as $\bar{O}.a_1 = A_Z$ and $\bar{U} = P_Z$, it comes out that

$$f_U(x) = \frac{A_Z}{P_Z} x.f_O(x)$$

Which implies (13) with proportionality coefficient of $A_Z / P_Z = 1/\bar{x}_O$. To sum up,

$$f_U(x) = \frac{1}{\bar{x}_O} x.f_O(x) \tag{14}$$

2.3.2. Relationship between the statistical moments of the two distributions

From (14) stems a generic relation between the statistical moments of the two distributions (Kleinrock, 1975): At any order r, it holds that

$$E_U[x^r] = \frac{1}{\bar{x}_O} E_O\left[x^{r+1}\right] \tag{15}$$

This relation corresponds to the zone-based formula in Stairs (1977). At order $r = 1$, the average density as experienced by the users satisfies that

$$\bar{x}_U = \frac{1}{\bar{x}_O} E_O\left[x^2\right] = \bar{x}_O(1+\gamma_O^2) \tag{16a}$$

It is equivalent to a ratio of the average densities (RADs):

$$RAD_{U|O} \equiv \frac{\bar{x}_U}{\bar{x}_O} = 1+\gamma_O^2 \tag{16b}$$

Both relations correspond to formulas established by Lewontin & Levins (1989) using zones as statistical units and Ottensmann (2018a) using land units. If the human density is homogenous in the territory, then $\gamma_O = 0$ and the average human densities according to either statistical population are equal. However, the larger the heterogeneity (as measured by the relative dispersion), the higher the ratio $\bar{x}_U / \bar{x}_O$ of used to offered average human densities.

Figure 1 illustrates both the used and offered PDFs. Its assumptions are the following: That x_O is distributed log-normal with parameters $m_O = 3.24$ (mean of $\ln(x_o)$) and $s_O = 1.76$ (standard deviation of $\ln(x_o)$), stemming from France-like conditions $\bar{x}_O = 120$ p/km² and $\gamma_O = 4.6$. The related x_u is log-normal, too, with parameters $m_U = 6.34$ and $s_U = s_O$, hence $\bar{x}_U = 2{,}661$ p/km² and $\gamma_U = 4.6$, too. In that particular instance, the ratio of average human densities, $\bar{x}_U / \bar{x}_O$, amounts to 22 – indeed a very large value.

2.3.3. Indicators of used human density heterogeneity

The indicators of heterogeneity recalled in §2.2.3 for the random variable of offered density also apply to that of used density. Its variance stems from moments of the offered density in a specific way: From (15) at order r=2,

$$E_U\left[x^2\right] = \frac{1}{\bar{x}_O} E_O\left[x^3\right]$$

Combining with (16a), it comes out that

$$V_U[x] = \frac{1}{\bar{x}_O} E_O\left[x^3\right] - \left(\frac{E_O\left[x^2\right]}{\bar{x}_O}\right)^2 \tag{17}$$

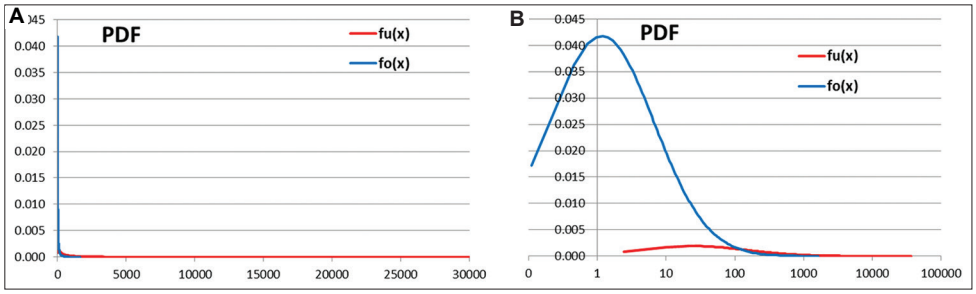

Figure 1. Used and offered PDFs of human density
(A) Standard scales, (B) abscissas in log-scale
Source: author's elaboration

When x_O follows a log-normal distribution, then so does x_U (cf. appendix C), with same variance s^2 of the law of ln x: Then, the interquartile ratios of x_U are identical to their offered counterparts.

2.4. Lorenz curve and Gini index

2.4.1. Recalling the classical theory of inequality measurement

In Gini's classical analysis of income inequalities (Gini, 1912; 1955; Gionanni & Gubbiotti, 2015), the sum of all individual incomes in a group of people is decomposed according to specific subgroups of people. A typical subgroup gathers the people that each earn less than a given level of income. Then, to the proportion $F_P(x)$ of-people whose income is $<x$ is associated the proportion $F_I(x)$ of total income that stems from the aggregation of their individual incomes. As both proportions are increasing functions of x, they depend on each other in a unique way. Their relationship is known as the "Lorenz function" denoted L and defined as follows (Cowell, 2009; Lorenz, 1905):

$$\alpha \mapsto L(\alpha) \equiv F_I \circ F_P^{(-1)}(\alpha)$$

The reason is that each value of x gives rise to population proportion $\alpha \equiv F_I(x)$, therefore satisfying $x = F_P^{(-1)}(\alpha)$, and to income proportion $F_I(x)$, which is thus equivalently expressed as $F_I(F_P^{(-1)}(\alpha))$.

The derivative $\dot{L}$ of L satisfies that

$$\dot{L}(\alpha) = \frac{f_I\left(x_P^{(\alpha)}\right)}{f_P\left(x_P^{(\alpha)}\right)} = \frac{1}{\bar{x}_P} x_P^{(\alpha)} \qquad (18)$$

It is non-negative and increasing with α since $F_P^{(-1)}$ is increasing: This makes L an increasing and convex function. Thus, in the diagram of F_I versus F_P in $[0,1]\times[0.1]$, the graph of function L, called the "Lorenz curve," lies below the straight line from point (0,0) to point (1,1) (Figure 2). The area between the straight line and the

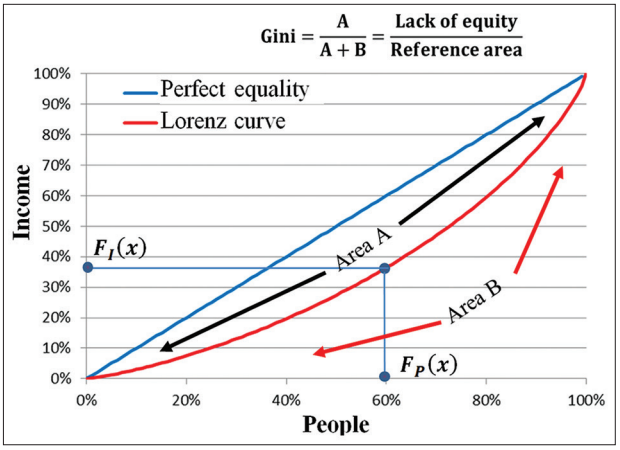

Figure 2. Lorenz function and Gini index
x designates an individual income
Source: Author's adaptation from https://en.wikipedia.org/wiki/Gini_coefficient [Last accessed 8.11.2022]

Lorenz curve, divided by the area below the straight line, that is, 0.5, is known as the Gini index, with mathematical formula as follows (Cowell, 2009):

$$G \equiv 2\int_0^1 (\alpha - L(\alpha))d\alpha \qquad (19)$$

The Gini index takes its value in $[0,1]$. Between different income distributions, the larger the heterogeneity, the larger the Gini index: It is a metric of inequality (Cowell, 2009). In appendix C, a log-normal instance is addressed to give insight in the consumption model and illustrate the properties of relative dispersions and the Gini index.

2.4.2. Adaptation to human density

Gini's line of reasoning applies to the distributions of human density: To the $F_O(x)$ share of space with density $<x$ corresponds the $F_U(x)$ share of people each experiencing individual density $<x$. Here, the Lorenz function is $L \equiv F_U \circ F_O^{(-1)}$. The resulting Gini index constitutes another metric of density heterogeneity, along with γ_O and γ_U. Figure 3 exhibits a Lorenz curve of population density,

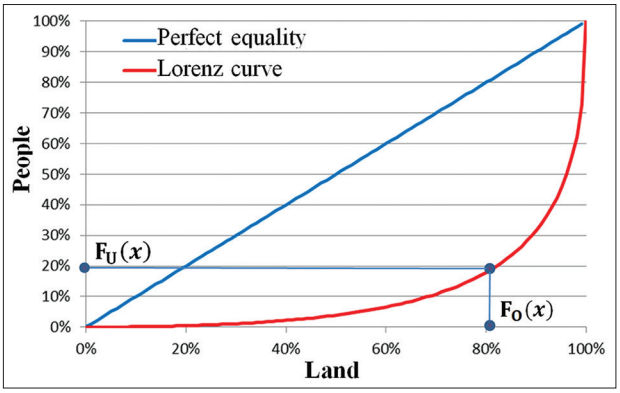

Figure 3. Lorenz function of human density
Source: Author's elaboration

assuming the same used and offered distributions of human density, as shown in Figure 1. This particular instance exhibits a Gini index of 0.79, which is a very high value for that kind of index (Cowell, 2009).

The Lorenz curve depicts the relation between land and people as it relates a proportion of people, on the vertical axis, to the proportion of land that accommodates them, on the horizontal axis. The relation pertains to the spatial density of human settlement: Both the land units (horizontal axis) and the individuals (vertical axis) are ranked in increasing order of density x. Given a specific value x of human density, the proportion $F_o(x)$ of land accommodates the proportion $F_U(x)$ of people, all of them at density lower than x. Conversely, the residual $1-F_O(x)$ share of land accommodates the residual $1-F_U(x)$ share of people, all of them at density greater than x. Thus, the point $(F_O(x), F_U(x))$ splits the diagram in two parts: Lower density space and people on the left side, higher density space and people on the right side. The average densities on the lower and higher sides are proportional to $\frac{F_U(x)}{F_O(x)}$ and $\frac{1-F_U(x)}{1-F_O(x)}$, respectively, with coefficient of $P_Z / A_Z = \bar{x}_O$.

The ratio of higher and lower average densities (RADs) is thus equal to

$$RAD_{H|L} \equiv \frac{1-F_U(x)}{F_U(x)} \frac{F_O(x)}{1-F_O(x)} \tag{20}$$

For instance, in Figure 3, it appears that about 20% of people are accommodated in 80% of space. The used offered ratio of $\frac{20\%}{80\%}$ on the lower side, compared to $\frac{80\%}{20\%}$ on the higher side, imply that the average density in the higher part is about 16 times that in the lower part.

2.5. On alternative average indicators of density

Stairs (1977) introduced generic weighting systems for averaging the population-weighted density and measuring the heterogeneity of human density. Denoting a weighting system as a function $x \mapsto w(x)$ of density level x, the associated average density is stated as follows:

$$x_w \equiv \frac{E_U[w]}{E_O[w]} \tag{21}$$

Stairs (1977) also considered "generalized population density" as the r-th order root of the ratio of moments at orders $k+r$ and k:

$$x_k^r \equiv \left(\frac{E_O\left[x^{k+r}\right]}{E_O\left[x^k\right]} \right)^{\frac{1}{r}} \tag{22}$$

Average lived density $\bar{x}_U$ is a particular instance associated to $(k,r) = (1,1)$.

It is shown in appendix C that, when the offered density follows a log-normal distribution with median M_o and relative dispersion γ_O, then this average indicator satisfies that,

$$x_k^r = M_O(1+\gamma_O^2)^{k+r/2} = M_O(\bar{x}_U / \bar{x}_O)^{k+r/2}$$

Therefore, it is basically a power function of the ratio of used and offered average densities, $\bar{x}_U / \bar{x}_O$.

Complementarily to the population-weighted arithmetic mean density, Craig (1984) also considered the logarithm of density as an indicator of the magnitude of density. He argued that magnitude-based density indicators would be especially relevant to assess the variations of local density over time by laying the emphasis on local significance, since the local meaning of a given change of density depends on the initial, local density level. Using our notation, the average indicator of density magnitude states as $E_U[\ln x]$ and gives rise to the geometric mean of used density,

$$\tilde{x} \equiv \exp E_U[\ln x] \tag{23}$$

Craig (1984) related the geometric mean to "the ideas of information gain and entropy" and insisted on its property of decomposability along spatial sub-divisions. It is also akin to the Theil index of heterogeneity (Cowell, 2009). When the offered density follows a log-normal distribution $LN(m,s^2)$, then the used density is log-normal, too, with identical relative dispersion γ (cf. appendix C) and the average log value $E_U[\ln x]$ is equal to $m+s^2$, that is, to

$\mathrm{E_O}\left[\ln x\right]+\frac{1}{2}\ln(1+\gamma^2)$. Therefore, the related geometric mean $\tilde{x}$ satisfies the following equalities which makes it a geometric midpoint between $\bar{x}_\mathrm{O}$ and $\bar{x}_\mathrm{U}$:

$$\tilde{x}=\exp(m+s^2)=\bar{x}_\mathrm{O}\exp\left(\frac{1}{2}s^2\right)=\bar{x}_\mathrm{O}\sqrt{1+\gamma^2}=\frac{\bar{x}_\mathrm{U}}{\sqrt{1+\gamma^2}} \quad (24)$$

Thus, based on the case of log-normal distributions, it may be conjectured that alternative indicators of average density do not yield much gain beyond considering the $(\bar{x}_\mathrm{O},\bar{x}_\mathrm{U})$ pair of average densities.

3. A case study of France as of 2019

3.1. The territory under study

Metropolitan France comprises about 34,750 municipalities called "communes" (INSEE, 2021a). We take them as zones in the country. The country area of about 543 thousand square km yields an average commune area of 15.6 km² (Aliaga *et al.*, 2015).

As of 2019, the French metropolitan population amounts to about 65 M inhabitants (INSEE, 2021b). Thus, the average commune population is 1800 people only and the overall spatial density of population is about 120 persons per km². Figure 4 exhibits the map of French communes colored according to density level in the way of either the INSEE (Aliaga *et al.*, 2015), the French National Institute for Economic Statistics, or Eurostat, the statistical body of the European Union (Eurostat, 2019). It shows that most of the country area has low population density.

3.2. Used density versus Offered density

To obtain the statistical distribution of used density, we made the following assumption: That each commune's population is distributed evenly in its area. Of course, this is only an approximation as large communes (meaning communes of large area) are likely to exhibit significant intra-communal heterogeneity of human settlement. Based on this assumption, we ranked the communes of increasing average density. According to the ranking, we calculated two cumulated variables: First the land area, second the population. By dividing the cumulated area up to commune z by the total country area, the $\mathrm{F_o}$ CDF is obtained at point x_z. Similarly, by dividing the cumulated population up to commune z by the total country area, the $\mathrm{F_u}$ CDF is obtained at point x_z.

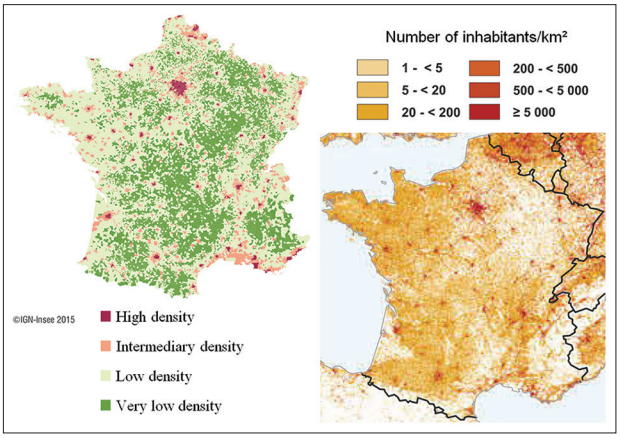

Figure 4. Human density of French communes as of 2015 and 2018
Sources: Aliaga *et al.* (2015), Eurostat (2019)

The next step is to draw the diagram of $\mathrm{F_u}$ versus $\mathrm{F_o}$, that is, the Lorenz curve (Figure 5A). The Gini index is easy to calculate, by accumulating $2\left(\mathrm{F_O}\left(x_z\right)-\mathrm{F_U}\left(x_z\right)\right).\left(\mathrm{F_O}\left(x_z\right)-\mathrm{F_O}\left(x_{z-1}\right)\right)$ over communes z. The outcome is 0.76, again a very large value. Furthermore, easy to calculate are the mean value, variance, standard deviation, and relative dispersion of the density variable either offered or used. The results are, in persons per square kilometer:

- for x_O: $\mathrm{E_O}\left[x\right]=120$ and $\mathrm{SD_O}\left[x\right]=548$, yielding $\gamma_\mathrm{O}=4.58$ (dimensionless).
- for x_U: $\mathrm{E_U}\left[x\right]=2{,}628$ and $\mathrm{SD_U}\left[x\right]=4{,}691$, yielding $\gamma_\mathrm{U}=1.79$ (dimensionless).

Figure 5B and C depicts the empirical CDFs $\mathrm{F_O}$ and $\mathrm{F_U}$, together with log-normal approximations that mimic the mean and variance of each distribution. To obtain PDFs (Figure 5D and E), we discretized the CDFs and derived the respective PDFs as the average value between two successive points. On trying to model the offered density as a lognormal distribution, a close match was obtained: Yet, a perfect lognormal model would entail identical relative dispersions between the offered and used distributions – a conclusion definitely not supported by the data. Looking for alternative conventional distributions to fit the data, Singh-Maddala CDFs were found appropriate (Figure 6).

More simply, it turned out that the used density CDF, $\mathrm{F_U}$, is about an affine linear function of Inx from its first to 8th deciles. A straightforward consequence is that, on the interval between the two deciles, $\breve{\mathrm{f}}_\mathrm{U}(x)\propto x^{-1}$. In turn, the offered density is about inverse quadratic, $\breve{\mathrm{f}}_\mathrm{O}(x)\propto x^{-2}$. Both approximations are well supported by the data (Figure 7). Some related analytical properties are provided in appendix D.

3.3. Decile values and the heterogeneity of human density

The decile values of the offered and used distributions of human density were derived from their respective

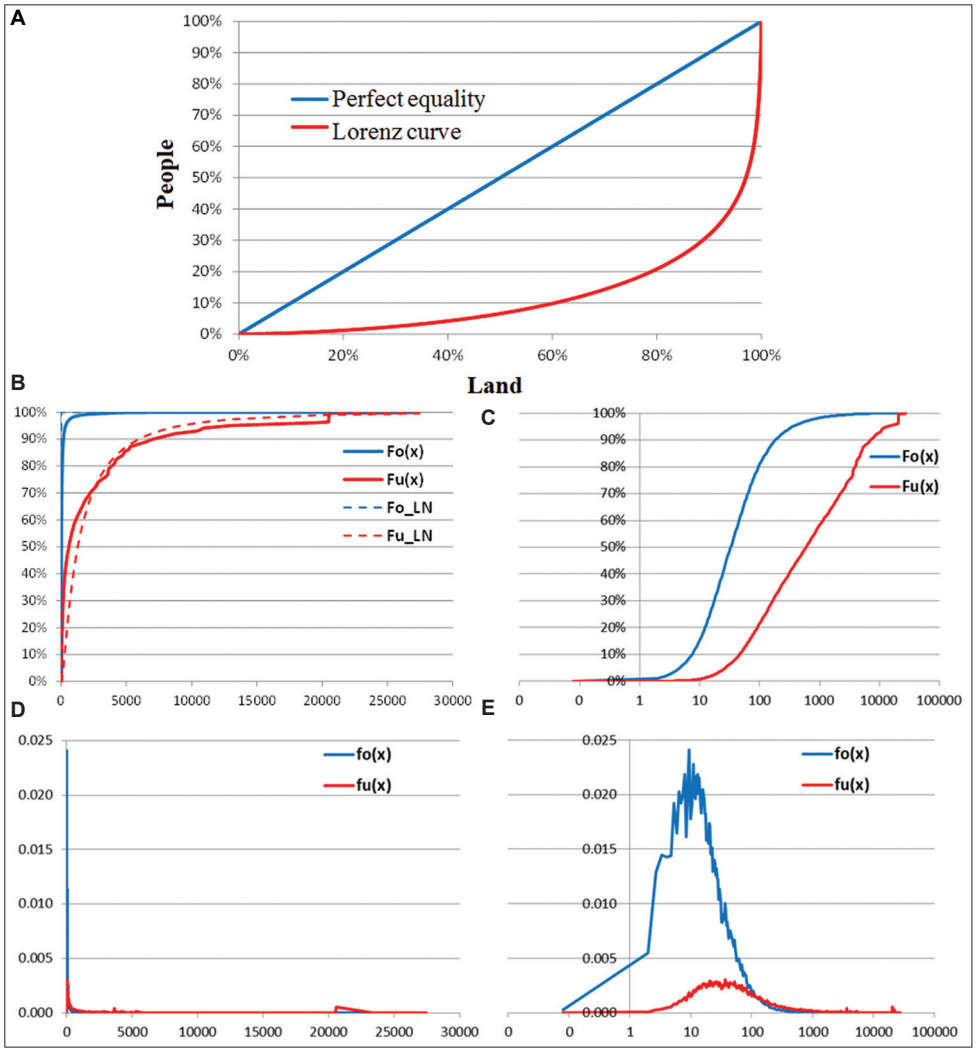

Figure 5. Human density in France, 2019
(A) Lorenz curve, (B) used and offered CDFs of human density, (C) same with abscissas in log-scale, (D) used and offered PDFs of human density, and (E) same with abscissas in log-scale
Source: Author's calculations based on INSEE data (2021a; 2021b).

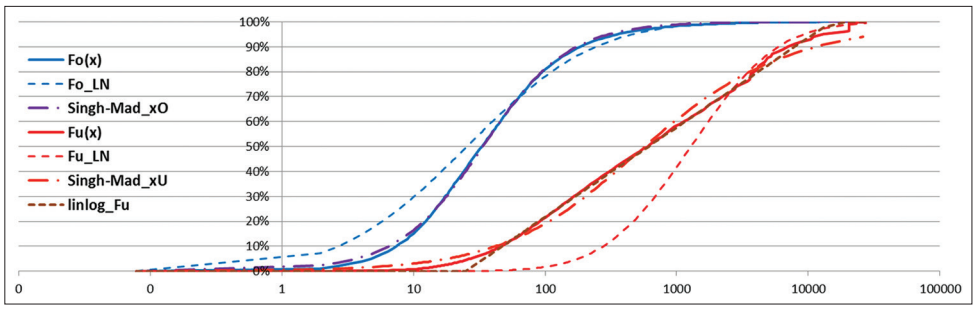

Figure 6. Used and offered CDF, with log-normal, Singh-Maddala, and linear-log approximations
Abscissas in log-scale
Source: Author's estimations based on INSEE data (2021a; 2021b)

CDF (Table 1). The median value of offered density is much lower than the offered mean – its significance is only to state that a major share of France's territory lies under very low density. The 80 – 20% shares of low versus.

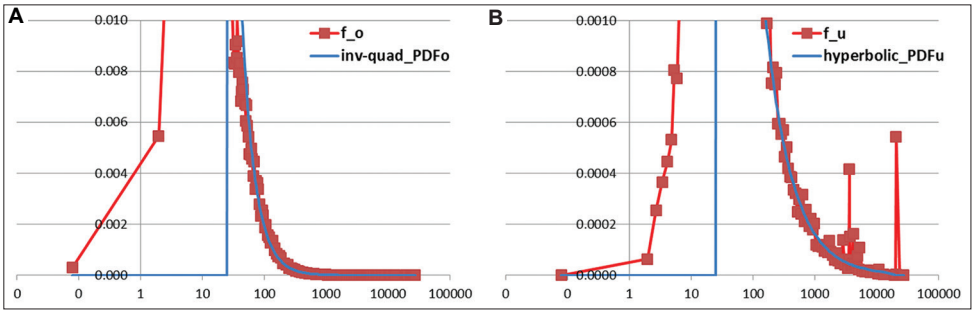

Figure 7. Approximations of (a) offered density PDF and (b) used density PDF
Abscissas in log-scale.
Source: Author's calculations based on INSEE data (2021a; 2021b).

Table 1. Deciles of offered density versus used density (France, 2019), in persons per km²

α	10%	20%	30%	40%	50%	60%	70%	80%	90%
$x_O(\alpha)$	7	12	17	24	33	46	64	96	186
$x_U(\alpha)$	46	92	167	306	600	1,146	2,213	3,872	6,999

Source: Author's calculations based on INSEE data (2021a; 2021b).

high density land are associated to 20 – 80% shares of low versus high density people. Such striking contrast calls for quantitative metrics to complement density maps in spatial analysis.

The deciles pave the way to the qualitative assessment of low to high levels of density. With respect to people living in France, the median used density, that is, 600 persons per square kilometer may be taken as "medium level of density," low densities for the bottom 20%, that is, below 92 p/km², high densities for the top 20%, that is, above 4000 p/km². These people-based values are close to the values selected by the Regional and Urban General Directorate of the European Commission (Eurostat, 2019). The land-based deciles have little relevance to depict urban conditions. The average offered density is just a ratio to summarize the intensity of human occupation over a given stretch of land – nothing less, nothing more, especially not about the used density of population.

The average densities are meaningful metrics. The standard deviation of offered density makes little sense to people: and not much more for land, in fact. The Gini index is much more meaningful and so are the relative dispersions of used and offered density.

As for interquartile ratios to measure distribution heterogeneity, the outcomes are:

- For offered density, inter-decile ratio (RID) of 186/7 = 27 and inter-quartile ratio (RIQ) of 80/15 = 5.3.
- For used density, inter-decile ratio (RID) of 7000/46= 152 and inter-quartile ratio (RIQ) of 3000/120 = 25.

The RID and RIQ of x_O are quite high for their kinds of indicators. As for x_U, the RID and RIQ values are still much higher: they reveal the very large heterogeneity of used density, that is, of human density as lived by the people.

We utilized the dataset to calculate the alternative indicators of average density recalled in §2.5. Craig's geometric mean of the used density has a value of $\tilde{x} =$ 590 p/km²: Thus, it is close to the geometric midpoint between $\bar{x}_O$ and $\bar{x}_U$, since $\tilde{x} \approx 4.45\bar{x}_O$ and $\bar{x}_U \approx 4.93\tilde{x}$. Both ratios are close to value $\sqrt{1+\gamma_O^2} = 4.69$ that was expected using a lognormal approximation of x_o.

The Stair's generalized indicators x_k^r in (22) were computed for indices k from 0 to 9 and r from 1 to 5. Figure 8 exhibits the reduced indicators $\ln(x_k^r / M_O)/(k+\frac{1}{2}r)$ as functions of k depending on r. The salient values are those associated to pairs $(k,r)=(0,1)$ and (1,1), that is, to $\bar{x}_O$ and $\bar{x}_U$, while the other pairs yield an overall pattern about value 1 that corresponds to lognormal distributions.

4. Discussion

4.1. On the statistical populations and random variables of human density

The history of statistics began with early censuses of the human population in a couple of countries, that is, with human populations as statistical populations – hence, the very name of the latter concept. In such a historical perspective, the used density lends itself to be modeled as a random variable in the human population. However, as geography and cartographic methods have been well developed long before the advent of computers, the plain, offered density has been introduced long before the used, lived density. In the pioneering contributions of Craig (1975; 1979; 1980; 1984) and Stairs (1977a), it has been clearly stated that the average plain density is area weighted, whereas the average lived density is a population-weighted

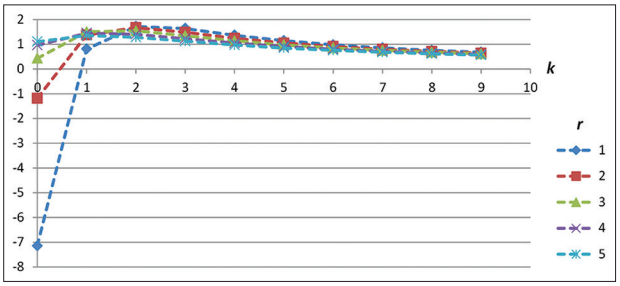

Figure 8. Stair's generalized density indicators under reduced form $\ln(x_k^r / M_O) / (k + \frac{1}{2} r)$

Source: author's calculations based on INSEE (2021a; 2021b)

one: This points to the respective statistical individuals. Lewontin & Levins (1989) went further in the probabilistic representation of human density by considering spatial cells of unit ground area as the statistical individuals in geographical space as a statistical population: The cell attribute of local human density is inherited by the people inhabiting the cells, therefore constituted the used density as an RV in the human population. Yet, the probabilistic framework of Ottensmann (2018a) is limited to the two statistical populations, the human density RVs on both sides and the relations between the average values.

In the present article, we introduced the respective PDFs of human density in either statistical population and we related them using a consumption model. This affiliation to (simple) probabilistic theory yields several benefits, from the mathematical statement of the PDF relationship which is a fundamental one, to the relation between statistical moments, and up to the consideration of the Lorenz function and the related Gini index. The relation between the respective averages of used versus offered density is a prominent instance of the relation between statistical moments. The reference to theory provides the general relation between moments in a straightforward way, compared to the former derivations of the relation between average densities (Lewontin & Levins, 1989; Ottensmann, 2018a). Stairs (1977a) had stated the relation between the statistical moments in a concise and general way, yet with no explicit consideration of PDFs.

In the application to France as of 2019, the PDFs of offered and used densities were easy to study on a standalone basis. Yet, to visualize both functions jointly, the standard diagram (Figure 5D) depicted mainly the quasi-disjunction of their supporting sets – lower values of x giving most of the probability weight to x_o versus much higher values giving significant probability weight to x_U. The recourse to log-scale on the x-axis (Figure 5E) was instrumental to exhibit the PDF values on the y-axis and enable for some visual comparison. In such a diagram, however, it is less easy to visualize that the area below the PDF curve amounts to 1. The CDF functions are easier to depict jointly than their PDF counterparts. Using conventional abscissas (Figure 5B), function F_U increases in a smooth way paved by the decile values, while function F_O increases in a one-shot way at low values of x, making the decile values hardly legible. The recourse to log-scale on the x-axis (Figure 5C) enables one to visualize the magnitude of the deciles. The graphical practicality of density magnitudes, that is, of the logarithms of the human density, provides another reason to utilize them, beside the point made by Craig (1984) that relative changes in local population densities are more significant than absolute ones to assess the variations over time of human density in a geographical space.

4.2. On land units and the heterogeneity of land-use

Ottensmann (2018a) studied and discussed the definition of spatial cells, in other words, the zoning system to consider for local density and its assignment to people according to their zones of residence. Craig (1984) studied the effects of three zoning systems on the values of the used density in Great Britain as of 1971: He stated that "both the means (of offered and used density) increase as the (spatial) units are progressively subdivided." This is a straightforward consequence of the law of total variance applied progressively to finer and finer subgroups. It emphasizes that the notion of used density strongly depends the underlying system of residence zones. This must be kept in mind in the consideration of any set of numerical values of used density indicators. Such sensitivity also pertains to any indicator of heterogeneity in offered densities, since the zoning sensitivity of the average used density, on the left side of (16a), comes from that of the relative dispersion of the offered density, on the right side of (16a). In fact, the zoning system to estimate both offered density heterogeneity and average used density has to satisfy a twofold condition on zone sizes: A trade-off between, on the one hand, zones small enough to capture the spatial heterogeneity finely and, on the other hand, zones large enough to grasp the "life basins" where people live.

4.3. On indicators of heterogeneity in human density

For positive real variables, the relative dispersion is a heterogeneity indicator derived from the standard deviation divided by the mean. The explicit modeling of the PDFs of human densities, that is, f_o and f_U, induces their respective CDFs f_o and f_U: From these stem, the quantiles of their respective distribution, and in turn robust statistics, including the median as a middle value and also interquartile ratios as measures of heterogeneity, including the inter-quartile ratio and the inter-decile one. Not only are the interquartile ratios simpler than the alternative

indicators of average density put forward by Craig (1984) and Stairs (1977a), but also they are more informative in the case of France as of 2019.

The Gini index constitutes a heterogeneity indicator of its own kind. It is a statistical summary of the full Lorenz curve, which is a more comprehensive tool but also more disaggregate to apprehend heterogeneity in both x_O and x_U. Some hints of that appeared in previous studies such as Aliaga *et al.* (2015) who mentioned that 35% of French people are living in even 90% of French communes: Up to the difference between spatial entities and land units, the mention is analogous to one point on the Lorenz curve. The full Lorenz curve contains much more information. In the 2019 France case, it was found that 80% of people live in 20% of space and conversely that the remaining 80% of space accommodate the remaining 20% of people. Thus, it constitutes yet another instance of the Pareto principle that arises in many distributions from incomes in populations to the sizes of firms. It is consistent with the linear-log model of the used density CDF, since this model makes the used density PDF a limit Pareto law and the offered density a truly Pareto law.

4.4. On the applications of used density

The local density of a place is a specific attribute of individual people living there. The notion of x_U and its probabilistic features from f_U to $\overline{x}_U$ and γ_U constitute a simple statistical model to analyze human population according to human spatial density.

Ottensmann (2018a) pointed out to the existing areas of application of the used density: (i) Primarily as "a descriptive measure of distribution, often in comparison with conventional density," (ii) urbanization patterns and their dynamic variations, including urban sprawl, (iii) mobility analysis relating residential and job densities to the modal share of transit modes of transportation (Barnes, 2001), (iv) agglomeration economies, relating used density to total urban factor productivity (Rappaport, 2008a) and to urban consumption amenities (Rappaport, 2008b), and (v) energy use and GHG emissions from household residential and travel patterns (Lee & Lee, 2014).

In recent years, the concept of population-weighted density has disseminated, notably through web online contributions showing its descriptive power (Bradford, 2008a-c; Florida, 2012; NENAD, 2021; Ottensmann, 2016) and above all in the academic literature of its various fields of applications: (i) Geographical analysis (Hanberry, 2022) including studies on the COVID epidemiology (Pascoal & Rocha, 2022), (ii) urbanization patterns (Townsend & Ellis-Young, 2018) and urban sprawl (Ottensmann, 2018b), (iii) mobility analysis (An *et al.*, 2022), (iv) economics (Albouy & Stewart, 2012; Faberman & Freedman, 2016; Krugman, 2013), and (v) environmental impacts (Huang & Brown, 2021) and potentials (Lu *et al.*, 2022). However, as of end 2022, population-weighted density still had not an entry of its own in the English-speaking Wikipedia, where "living density" is just mentioned as an alternative measure of human density (Wikipedia, 2022).

4.5. Further developments

The notion of population-weighted density was introduced in the mid-1970s by a demographer (Craig) and a chemistry scientist (Stairs), perhaps because quantitative socioeconomic analysis is a science of composition as is chemistry. Since then it has been adopted by geographers, economists interested in regional science and urban economics, and transport scientists.

Lived density may well be viewed as a simple form of spatial accessibility, as theorized by Hansen (1959), Poulit (1974) and Koenig (1974; 1980): The Hansen accessibility index, taken at a given zone as origin of trip-making, aggregates opportunities of a given kind over a larger territory, with numbers weighted by a declining function of origin-destination distance – or travel time or cost in the Poulit-Koenig formulation. This indicator has taken a central position in agglomeration economics (Fujita & Thisse, 2002) and geography economics (Krugman, 1997; Fujita *et al.*, 1999); it is also called the effective density in economic geography (Graham & Gibbons, 2019). It still remains to be considered as a property of the people residing in the origin zone, for all origin zones and all people, and to be analyzed as a random variable among the statistical population of people.

5. Concluding Remarks

The gist of the article is to model human density in geographical space using basic probability theory: (i) Statistical populations of land units and of people, respectively, (ii) putting human density as a random variable in both statistical populations, with specific PDFs and CDFs, (iii) relating the used density PDF to the offered density PDF through the probabilistic consumption model, (iv) deriving the statistical moments of the used density from those of the offered density, and (v) considering the Lorenz curve and the Gini index. The original contribution is 3-fold: The formal statement as random variables, the identification of the consumption model, and the consideration of heterogeneity indicators for human density in space (interquartile ratios and Gini index).

All of the concepts are well established in their own field, geography, or probabilistic modeling: The article provides a fresh perspective to relate the two fields – casting one

more bridge between them. The explicit consideration of used density enables for better understanding the spatial occupation. The progress to harvest is the same one as in other instances of consumption models: Not only (i) Gini's analysis of income inequality, but also (ii) the queuing theory of waiting times (e.g., Kleinrock, 1975), (iii) Wardrop's model of temporal vs. spatial distributions of vehicular speeds on roadways (Wardrop, 1952), and (iv) a model of transit vehicle loads and transit users' exposure to crowding conditions (Leurent *et al*., 2012; 2017).

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Appendix

Appendix A: Nomenclature

z zone, a spatial entity in set Z covering the territory under study

A_z, ground area of zone z

P_z, population in zone z

A_z, overall ground area of territory

P_z, overall population in territory

a_1, unit ground area

o a land unit

P_o, population in o

$\bar{O}$, total number of land units (equal to A_Z / a_1)

$\bar{U}$, total number of people in territory (equal to P_Z)

x, density level

f_O & F_O, PDF & CDF of x regarding land units, with mean $\bar{x}_O$ and relative dispersion γ_O

f_U & F_U, PDF & CDF of x regarding people, with mean $\bar{x}_U$ and relative dispersion γ_U

L, Lorenz function

G, Gini index

Appendix B: Consumption model

A consumption model can be stated in a generic fashion as follows. It relies on a consumption function say $c : x \mapsto c(x)$, which takes nonnegative real values and measures the amount of consumption made by an individual with attribute x.

Let then f_O denote the PDF of attribute x in the statistical population of such individuals. The consumed units of all the individuals make up a statistical population of their own, with PDF function f_U that satisfies the following relation:

$$f_U(x) \propto c(x).f_O(x) \tag{B-1}$$

Postulating that the consumption function is monotonous, then eqn. (B-1) can be demonstrated using the same proof as for Equation (13). The proportionality coefficient is the reciprocal of $\bar{c}_O \equiv \int c(x) f_O(x) dx$. Thus

$$f_U(x) = \frac{1}{\bar{c}_O} c(x).f_O(x) \tag{B-2}$$

Appendix C: Log-normal distributions and their basic properties

The log-normal distribution

The log-normal distribution is especially well-suited to consumption models of two kinds: Power laws, on the first hand (e.g., Cowell, 2009), and log-normal CDFs, on the other hand. The latter kind has been used by Cramer (1962) to study the diffusion of car motorization among a population of households. Here, we shall focus on the former kind, with some power r that needs not be an integer:

$$c(x) = c_1.x^r \tag{C-1}$$

Of course, factor c_1 needs be nonnegative to make some sense.

Basic properties of log-normal distributions

Let us recall the definition of a unidimensional log-normal distribution: A real random variable X is said to be distributed $LN(m,s^2)$ if it is positive and its natural logarithm is Gaussian, that is, $\ln(X) \sim N(m,s^2)$. Denoting as Φ the CDF of a reduced Gaussian variable and $\varphi(t) \equiv \exp(-t^2/2)/\sqrt{2\pi}$ the associated PDF, and letting $t_x \equiv (\ln(x) - m)/s$, the following outcomes are derived successively in a straightforward way (e.g., Cowell, 2009):

$$F_O(x) = \Phi(t_x)$$

$$F_O^{(-1)}(\alpha) = \exp(m + s.\Phi^{(-1)}(\alpha))$$

$$f_O(x) = \frac{1}{s.x}\varphi(t_x)$$

$$E_O[x] = \exp(m + \frac{1}{2}s^2)$$

$$V_O[x] = (E_O[x])^2 (\exp(s^2) - 1)$$

$$\gamma_O = \sqrt{\exp(s^2) - 1}$$

Hence $s = \sqrt{\ln(1 + \gamma_O^2)}$.

Furthermore, any derived random variable $Y \equiv c_1.X^r$ with $c_1 > 0$ is a log-normal variable, too. This is because $Y \geq 0$ and $\ln(Y) = \ln(c_1) + r.\ln(X)$, implying that $\ln(Y) \sim N(\ln(c_1) + r.m, (rs)^2)$, making Y an LN variable with parameters $\ln(c_1) + r.m$ and $(rs)^2$.

Moment formulas for log-normal distributions

The "Truncated Moments" formula

Coming to the population of consumed units in a consumption model with power function, we can avail ourselves of the "truncated moment" formula, namely:

$$\int_a^b x^r dF_O(x) = e^{r(m+rs^2/2)} \left\{ \Phi\left(\frac{\ln(b)-m}{s} - rs \right) - \Phi\left(\frac{\ln(a)-m}{s} - rs \right) \right\} \qquad \text{(C-2)}$$

An immediate consequence is that

$$E_O\left[x^r\right] = \exp(rm + \frac{1}{2} r^2 s^2)$$

Proof of (C-2). The reason is that

$$\int_a^b x^r dF_O(x) = \int_{t_a}^{t_b} e^{r(m+st)} \varphi(t) dt = e^{r(m+rs^2/2)} \int_{t_a}^{t_b} \varphi(t-rs) dt$$

in which we replace $\int_{t_a}^{t_b} \varphi(t-rs) dt$ with $\Phi(t_b - rs) - \Phi(t_a - rs)$.

It follows that $F_U(x) = \Phi(\frac{1}{s}(\ln(x) - m) - rs)$, i.e., that, in the population of consumed units, level x is distributed $LN(m+rs^2, s^2)$.

Thus $E_U[x] = \exp(m + rs^2 + \frac{1}{2}s^2)$ and $\gamma_U = \sqrt{e^{s^2} - 1} = \gamma_O$.

The case of r = 1

When $r = 1$, $F_U(x) = \Phi(\frac{1}{s}(\ln(x) - m) - s)$ and $E_U[x] = \exp(m + \frac{3}{2}s^2)$.

It is then easy to obtain

$$\frac{\bar{x}_U}{\bar{x}_O} = 1 + \gamma_O^2 = \exp(s^2) \qquad \text{(C-3)}$$

On Stairs' generalized population density

If the offered density is distributed $x_O \sim LN(m, s^2)$ then we have that

$$E_O\left[x^k\right] = \exp(km + \frac{1}{2}k^2 s^2)$$ and similarly

$$E_O\left[x^{k+r}\right] = \exp((k+r)m + \frac{1}{2}(k+r)^2 s^2)$$

Yielding that

$$\frac{E_O\left[x^{k+r}\right]}{E_O\left[x^k\right]} = \exp(rm + \frac{1}{2}(r^2 + 2rk)s^2)$$

And in turn, a Stairs' generalized density indicator of

$$x_k^r \equiv (\frac{E_O\left[x^{k+r}\right]}{E_O\left[x^k\right]})^{1/r} = \exp(m + \frac{1}{2}s^2(r+2k)$$

$$= M_O(\bar{x}_U / \bar{x}_O)^{k+r/2}$$

since $\exp(m)$ is the median M_O of x_O and $s^2 = 1 + \gamma_O^2$.

Lorenz curve and Gini index

Here, the Lorenz function, $L \equiv F_U \circ F_O^{(-1)}$, involves $F_O^{(-1)}(\alpha) = \exp(m + s.\Phi^{(-1)}(\alpha))$ together with $F_U(x) = \Phi(\frac{1}{s}(\ln(x) - m) - s)$. It thus is a function of α parameterized by *s*:

$$L_s(\alpha) = \Phi(\Phi^{(-1)}(\alpha) - s) \qquad \text{(C-4)}$$

The Gini index, $G_s \equiv 2\int_0^1 (\alpha - L_s(\alpha)) d\alpha$, can be considered as a function of *s*. It holds that

$$G(s) = 2\Phi\left(\frac{s}{\sqrt{2}}\right) - 1 \qquad \text{(C-5)}$$

Proof of (C-5). At point $s = 0$, as $\Phi \circ \Phi^{(-1)}(\alpha) = \alpha$, then $G_0 = 2\int_0^1 (\alpha - \alpha) d\alpha = 0$.

Differentiating G_s with respect to *s*, we get that:

$$\dot{G}(s) \equiv \frac{dG_s}{ds} = 2\int_0^1 \varphi(\Phi^{(-1)}(\alpha) - s) d\alpha$$

Changing variables according to $t \equiv \Phi^{(-1)}(\alpha)$ hence $d\alpha = \varphi(t) dt$, we get that

$$\dot{G}(s) = 2\int_{-\infty}^{+\infty} \varphi(t-s).\varphi(t) dt$$

Rearranging

$$(t-s)^2 + t^2 = 2t^2 - 2ts + s^2 = 2\left(t - \frac{1}{2}s\right)^2 + \frac{1}{2}s^2$$

$$= (\sqrt{2}t - \frac{s}{\sqrt{2}})^2 + (\frac{s}{\sqrt{2}})^2$$

It follows that $\varphi(t-s).\varphi(t)=\varphi\left(\sqrt{2}t-\frac{s}{\sqrt{2}}\right).\varphi(\frac{s}{\sqrt{2}})$ and in turn that

$$\dot{G}(s)=2\varphi\left(\frac{s}{\sqrt{2}}\right)\int_{-\infty}^{+\infty}\varphi\left(\sqrt{2}t-\frac{s}{\sqrt{2}}\right)dt$$
$$=\sqrt{2}\varphi\left(\frac{s}{\sqrt{2}}\right)\int_{-\infty}^{+\infty}\varphi(u)\,du$$
$$=\sqrt{2}\varphi(\frac{s}{\sqrt{2}})$$

By integration,

$$\int_0^s\dot{G}(v)\,dv=\sqrt{2}\int_0^s\varphi\left(\frac{v}{\sqrt{2}}\right)dv=2\int_0^{\frac{s}{\sqrt{2}}}\varphi(w)\,dw$$
$$=2\left[\Phi\left(\frac{s}{\sqrt{2}}\right)-\Phi(0)\right]$$

Lastly, formula (C-5) is obtained on making use of: $G(s)=G_0+\int_0^s\dot{G}(v)dv$.

Appendix D: Linear-log model of used density CDF

Here, it is assumed that, on a range $[A,B]$ of x, the used density CDF, F_u, is an affine linear function of ln x: thus, for $x\in[A,B]$,

$$F_U(x)=F_U(A)+\frac{F_U(B)-F_U(A)}{\ln B-\ln A}(\ln x-\ln A)\,. \qquad \text{(D-1)}$$

Differentiating with respect to x, the used density PDF is obtained as a hyperbolic function:

$$f_U(x)=\frac{1}{x}\frac{F_U(B)-F_U(A)}{\ln B-\ln A}\ \text{ for }\ x\in[A,B]\,.$$

From the general relationship (14), the offered density is determined as

$$f_O(x)=\frac{\bar{x}_O}{x^2}\frac{F_U(B)-F_U(A)}{\ln B-\ln A}\ \text{ for }\ x\in[A,B]\,.$$

From this stem the offered density CDF on the range:

$$F_O(x)=F_O(A)+(\frac{1}{A}-\frac{1}{x})\bar{x}_O\frac{F_U(B)-F_U(A)}{\ln B-\ln A} \qquad \text{(D-2)}$$

On the [A,B] range, formulas (D-1) and (D-2) enable one to recover the quantiles of x_U and x_O.

For $\alpha\in[F_U(A),F_U(B)]$, the quantile $x_U^{[\alpha]}$ of x_U at order α satisfies that

$$\frac{\alpha-F_U(A)}{F_U(B)-F_U(A)}=\frac{\ln x-\ln A}{\ln B-\ln A}\text{, therefore}$$
$$x_U^{[\alpha]}=A(\frac{B}{A})^{\frac{\alpha-F_U(A)}{F_U(B)-F_U(A)}}$$

The quantile $x_O^{[\alpha]}$ of x_O at order $\alpha\in[F_O(A),F_O(B)]$ satisfies that

$$\frac{\alpha-F_O(A)}{F_U(B)-F_U(A)}=\left(\frac{1}{A}-\frac{1}{x}\right)\frac{\bar{x}_O}{\ln B-\ln A}\text{, therefore}$$
$$x_O^{[\alpha]}=1/\left(\frac{1}{A}-\frac{\ln B-\ln A}{\bar{x}_O}\frac{\alpha-F_O(A)}{F_U(B)-F_U(A)}\right)$$

If the linear-log assumption is valid on the full range of x_U, then $F_U(A)=0$ and $F_U(B)=1$, yielding that

$$x_U^{[\alpha]}=A(\frac{B}{A})^{\alpha}=(A)^{1-\alpha}(B)^{\alpha} \qquad \text{(D-3)}$$
$$\bar{x}_U=\frac{B-A}{\ln B-\ln A}$$

From $F_O(A)=0$ and $F_O(B)=1$, $\bar{x}_O=\frac{\ln B-\ln A}{\frac{1}{A}-\frac{1}{B}}$

$$\frac{1}{x_O^{[\alpha]}}=\frac{1-\alpha}{A}+\frac{\alpha}{B}\,.$$

Applying (D-3) to order $\alpha=\frac{1}{2}$, the median M_U of x_U satisfies $M_U=\sqrt{AB}$. Applying it again to orders ¾ and ¼, the interquartile ratio RIQ_U satisfies $RIQ_U=\frac{x_U^{[0.75]}}{x_U^{[0.25]}}=\sqrt{B/A}$. From the empirical M_U and RIQ_U, the A and B parameters for France as of 2019 are recovered as

$$A=\frac{M_U}{RIQ_U}=\ 24\ \text{p/km}^2,$$

$$B=M_U RIQ_U=\ 14{,}895\ \text{p/km}^2.$$

ACCSCIENCE PUBLISHING

International Journal of Population Studies

RESEARCH ARTICLE

Levels and trends estimate of sex ratio at birth for seven provinces of Pakistan from 1980 to 2020 with scenario-based probabilistic projections of missing female birth to 2050: A Bayesian modeling approach

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Abstract

Most evidence on son preference in Pakistan is reflected in the higher child mortality among females than males. The sex discrimination before birth is rarely reported in Pakistan. This is the first study to quantify prenatal sex discrimination in Pakistan on a subnational level. We provide annual estimates of the sex ratio at birth (SRB) from 1980 to 2020 and scenario-based projections of the number of missing female births up to 2050 by Pakistan province. The results are based on a comprehensive database consisting of 832,091 birth records from all available surveys and censuses. We adopted a Bayesian hierarchical time series model to synthesize different data sources. We identified Balochistan with an existing imbalanced SRB since 1980. For the rest provinces without past or ongoing SRB inflation, we projected the largest female birth deficit to occur in Punjab in 2033 under the scenario that the SRB transition process starts in 2021. We demonstrated important disparities in the occurrence and quantification of missing female births up to 2050.

Keywords: Bayesian hierarchical model; Pakistan; Scenario-based projection; Sex ratio at birth; Son preference; Sex-selective abortion; Subnational modeling; Time series models

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1. Introduction

The ratio of the number of male live births to the number of female live birth, namely, the sex ratio at birth (SRB), is an essential element in estimating and projecting population size and its dynamics. Furthermore, an imbalanced SRB in a population reflects discrimination and disadvantages that females face in social, political, economic, and cultural contexts (Gupta *et al.*, 2003; Guilmoto, 2009). SRBs are distorted from their natural levels in several countries, primarily clustered in South Asia, East Asia, and East Europe. The imbalanced values reach as high as 1.2 (i.e., 120 male births per 100 female births) in certain regions (Attané & Guilmoto, 2007; Chao *et al.*, 2019a; Duthé *et al.*, 2012; Goodkind, 2011; Guilmoto *et al.*, 2009; Guilmoto, 2012; Guilmoto & Ren, 2011; Lin, 2009; Park & Cho, 1995). SRB is the

prime indicator of prenatal sex discrimination and sex imbalance in human populations.

The objectives of this study are (i) to provide annual estimates of SRBs among seven provinces of Pakistan from 1980 to 2020, (ii) to provide scenario-based projections to 2050 using a reproducible Bayesian statistical model, and (iii) to identify provinces with SRB imbalance. Our study has several contributions as a result of achieving the research objectives. First, to the best of our knowledge, this is the first study on Pakistan SRB that has produced provincial estimates and projections during 1980 – 2050. Second, it is the 1st time Balochistan is identified with the existence and transition process of the sex ratio imbalance using a Bayesian model. Third, based on the SRB imbalance results of the Bayesian hierarchical time series model, we compute the number of missing female births over time in provinces with imbalanced SRB and quantify the female birth deficits in each Pakistan province. Our study included seven provinces of Pakistan: Balochistan, Khyber Pakhtunkhwa, Punjab, Sindh, Gilgit Baltistan, Islamabad (ICT), and Azad Jammu and Kashmir. The results for Federally Administered Tribal Areas are omitted because of the unavailability of the longer time series data on SRB.

The remainder of this paper is organized as follows. Section 1.1 provides the theoretical background of the study. Section 2.1 summarizes the database compiled for statistical modeling and Section 2.2 summarizes the Bayesian statistical model used for provincial SRB estimation and the post-modeling process (identifying provinces with imbalanced SRB and calculating the number of missing female births). Section 3 presents the SRB results by province, the provincial SRB imbalances, the corresponding missing female births, and the scenario-based missing female birth projections. Sections 4 and 5 summarize the primary contributions and limitations and conclude the study.

1.1. Theoretical background

Distortion in the SRB has been primarily attributed to three interlinked factors (Guilmoto, 2009; 2012): (1) Son preference, (2) technological advances in prenatal diagnosis, and (3) preferences for smaller family size and consequent fertility decline. In countries with a patrilineal culture and shrinking family size, when prenatal sex determination and abortion technology are available, couples practice sex-selective abortion to secure at least one son. The SRB in such populations is male biased. SRB imbalance has been reported in 12 countries/areas since 1970 (Chao *et al.*, 2019a).

Pakistan is a country that has a strong preference for sons (Atif *et al.*, 2016; Hussain *et al.*, 2000; Khan & Siragelidin, 1977; Sathar *et al.*, 2015). Preference for male births in Pakistan stems from lineage, economic and social conditions, caste, and identity. At least one son in a strongly patrilineal society is essential for living arrangements in old age. One study suggested that the ideal family size in Pakistan (four children) has remained constant since the 1970s; moreover, the ideal sex composition of the children is more than 1 son (Wazir & Shaheen, 2016). Son preference is evidenced by the excess mortality of female children over male children under five in Pakistan, indicating possible differential treatment between girls and boys in this age group (Alkema *et al.*, 2014; Sathar *et al.*, 2015). The education attainment gap between females and males is large in Pakistan. Between 2017 and 2018, 30% of young women (age 15 – 24) completed middle or higher education compared to 50% of young men (National Institute of Population Studies (NIPS) [Pakistan] & ICF, 2019). Between 2018 and 2019, 36% of girls (ages 5 – 16) were out of school versus 25% of boys (Pakistan Bureau of Statistics (PBS) 2019). However, little evidence of prenatal sex preference has been reported in Pakistan. The previous studies identified no imbalanced SRB at the national level (Zaidi & Morgan, 2016; Chao *et al.*, 2019a). Other studies suggested that, among couples in Pakistan, the desire for a large family might dominate preferences for children of a particular type (De Tray, 1984). A high prevalence of sex-selective abortion was identified in two rural districts in Balochistan province (Qayyum & Rehan, 2017). However, the results mentioned above are based on survey data with small sample sizes.

The national scale levels and trends in SRB can mask the disparity among subregions in a country. Even in countries such as China and India, with an overall strong preference for sons, the SRB is not imbalanced in every province or state (Chao & Yadav, 2019; Chao *et al.*, 2020; Ge *et al.*, 2020; Jiang & Zhang, 2021). In Pakistan, a subnational level assessment of SRB is essential because the demography, socioeconomic status, and cultures (i.e., caste and ethnicity) are considerably heterogeneous. The latest estimates from the Pakistan Demographic and Health Survey (DHS) 2017 – 2018 revealed a high heterogeneous SRB across provinces: High inflation at 1.16 in Balochistan, a roughly normal SRB in Punjab at 1.05, and a female bias in Sindh and Khyber Pakhtunkhwa (SRB is 0.91 and 0.95, respectively) (National Institute of Population Studies (NIPS) [Pakistan] & ICF, 2019). To the best of our knowledge, no study has provided the annual estimates of the provincial SRB in Pakistan using all available data since 1980. To accurately determine whether the SRB is imbalanced in Pakistan and if so, where the imbalance occurs, it is essential to estimate the SRB on the subnational level.

Estimating the SRB in Pakistan is challenging for two reasons. First, limited data are available on birth histories in the past. Without a fully developed vital registration system in Pakistan, administrative birth records are lacking, and vital events are mostly estimated based on household surveys. Only a few sample surveys provided birth histories over different periods since the 1990s. Second, the data quality of census counts is typically low because of age heaping (Feeney & Alam, 1998). In historical census data, the number of children ever born in Pakistan is either unavailable or is unreliably reported. For example, birth histories were not collected in the 1981 Pakistan census (Ali *et al.*, 2001). Accordingly, the individual-level data of the three most recent censuses in Pakistan (conducted in 1981, 1998, and 2017) contain only the populations of boys and girls under 1 year old. The SRB data from sample surveys such as Pakistan DHS are suffering from large uncertainties because of the small sample sizes and misreporting of female births.

When estimating and projecting the provincial SRB in Pakistan, it is crucial to assess the levels and trends in the SRB by a reproducible statistical model. Using a Bayesian modeling approach for estimation and projection, observations from different data sources with varying levels of uncertainties can be synthesized and pooled together in a systematic and reproducible fashion. The Bayesian method can take into account both provincial SRB observations and external information on the SRB imbalance process to assist in model estimation and projection.

2. Data and methods

2.1. Data sources

Table 1 summarizes our database of provincial SRBs in Pakistan, with 531 SRB observations available in eight provinces of Pakistan. The reference years of these observations range from 1965 to 2019. The database contains 832,091 birth records by summing up the number of birth records where available. The number of birth records is unknown in some data series. Hence, the actual number of birth records involved in the study is more than what we reported here. The SRB observations were generated from the individual birth records in data sources with full birth histories (appendix for details of the data processing steps). The database is available as Supplementary File 1 (https://doi.org/10.6084/m9.figshare.21548082).

The DHS (ICF International, 2022) and Multiple Indicator Cluster Survey (MICS) provide the birth histories (either the full birth histories or the birth histories during the past 24 months before the survey interview) of women interviewed in retrospective survey questionnaires. Birth records are excluded if they were born more than 20 years prior to the year in which surveys were conducted to minimize recall bias from older women. Furthermore, the Pakistan Social and Living Standards Measurement Survey (PSLM) is a provincial-level survey with high coverage of households in Pakistan (Pakistan Bureau of Statistics [PBS], 2019). The PSLM records births over the 12 months before the date of the survey interview. The census is conducted once per decade and collects births in the 12 months preceding the census (Minnesota Population Center, 2019).

Given Pakistan's lack of reliable administrative birth data, it is essential to include all available data from different surveys to produce more reliable estimates and projections. The practice of making use of data from multiple data sources in estimation and projection has been widely used by international agencies, including the UNICEF, UN Population Division, and the Global Burden of Disease, and researchers in global and public health to reduce systematic bias from a single data source, to increase the length of the period that is covered by data (Alkema *et al.*, 2016; Bearak *et al.*, 2018; Gerland *et al.*, 2014; Liu & Raftery, 2020; Masquelier *et al.*, 2018; Wang *et al.*, 2020; You *et al.*, 2015). The data sources, we used as listed in Table 1, are based on provincial representative samples. If any future in-depth survey-specific consistency checks provide concrete evidence of bias in the examples of certain data sources, that particular data source should not be included

Table 1. Pakistan provincial SRB database

Survey name	Sample design	Survey year	# SRB observations	# Births records
Census	Census enumeration	1973, 1981, 1998	15	424,739
DHS	Two-stage stratified sample design	1990 – 1991, 2006 – 2007, 2012 – 2013, 2017 – 2018, 2019	301	253,580
MICS	Two-stage stratified sample design	2010, 2011, 2014, 2003 – 2004, 2007 – 2008, 2016 – 2017, 2017 – 2018	37	153,772†
PSLM	Two-stage stratified sample design	1995 – 2016, 2005 – 2006, 2007 – 2008, 2013 – 2014, 2018 – 2019	228	–
Total			531	832,091*

Note: DHS: Demographic and Health Survey; MICS: Multiple Indicator Cluster Survey; PSLM: Pakistan Social and Living Standards Measurement Survey. †: Number of birth records available only in MICS 2017 – 2018; –: The number of birth records is unavailable; *: The total number of births obtained by summing the available number of birth records in the 20 years before the survey conducted.

in the modeling. As of now, no evidence of biased sampling has been reported for any of these data sources. With their own objectives of the survey, these data sources provide a wealth of information, and there is no supporting evidence to choose one at the cost of the others. Hence, we use all these sources in our model estimation and projection.

Figure 1 illustrates the large uncertainty in SRB observations on national and provincial levels. It shows the observed SRB from Pakistan 2012 to 2013 and 2017 to 2018 DHSs on the national and provincial levels. In the data series Pakistan DHS 2012 – 2013, the national SRB observations range from 0.935 in the year 1989 to 1.195 in the year 2003. The average sampling error associated with these observations is 0.073, equivalent to 7% of the coefficient of variation (CV; refers to the ratio of sampling error to the value of observations, indicating the extent of variability given the observation value (Brown, 1998). On the provincial level, the SRB observations have a wider range with greater uncertainties because of smaller sample sizes. The smallest provincial SRB observation for DHS 2012 – 2013 is 0.917 in Islamabad (ICT) in year 2002 and the biggest value is 1.416 in Balochistan in year 1989. The average sampling error of all the provincial observations for DHS 2012 – 2013 is 0.087, corresponding to 8% of CV. Similarly, for Pakistan DHS 2017 – 2018, the observed SRB on national and provincial levels ranges from 0.914 to 1.178 and from 0.891 to 1.272, respectively. The average sampling errors are 0.087 and 0.139 for national and provincial data, respectively.

2.2. Methods

The model performance and predictive power were assessed by an out-of-sample validation exercise (leaving out recent

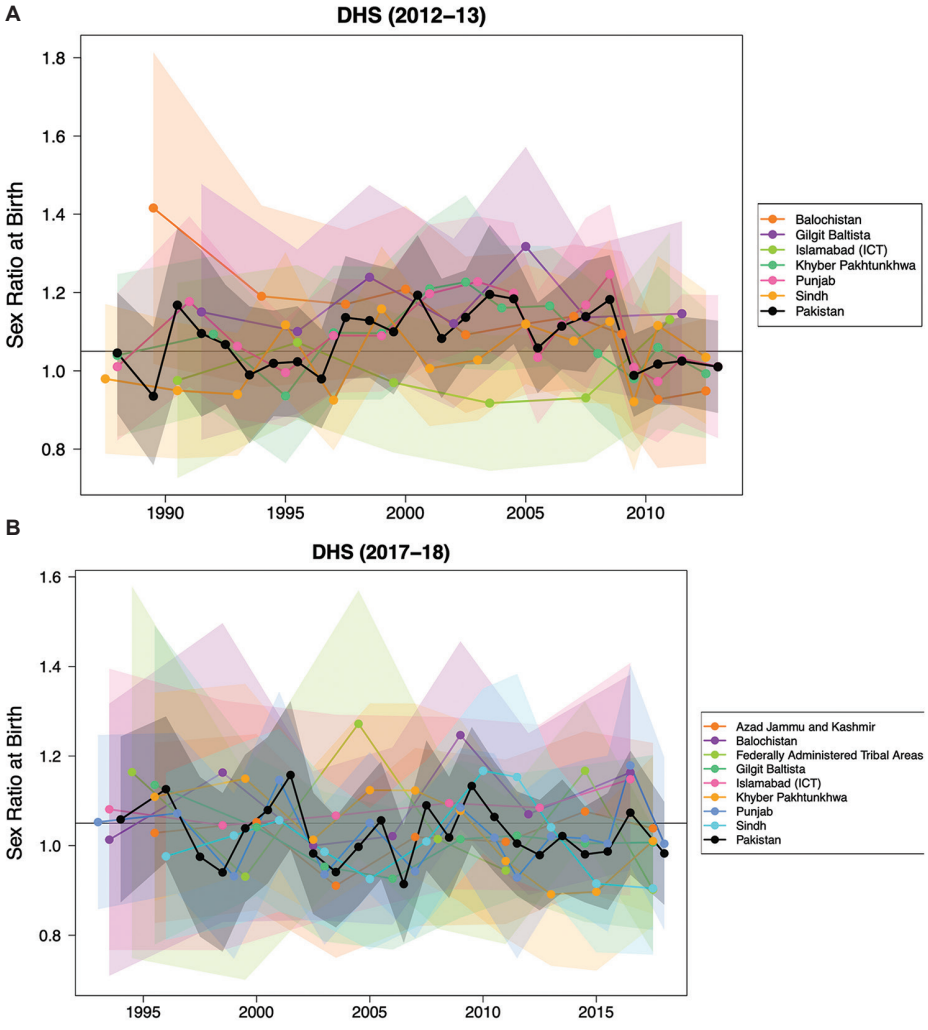

Figure 1. SRB data in (A) 2012 – 2013 and (B) 2017 – 2018 DHS on national and provincial levels
Note: The SRB observations (dots) from the same area (province or the whole of Pakistan) are connected with line segments of the same color. Shaded areas around the observation series represent the sampling errors in the series (quantified by twice the sampling standard errors).

observations) and simulation exercises (appendix for details). The validation and simulation results suggest good calibration and predictive power of the model.

The remainder of this section overviews the SRB Bayesian model.

2.2.1. Bayesian model for provincial SRB estimation and projection

The model for the SRB in Pakistan province is based on the model described previously by Chao *et al.* (2021a, b). In this study, we made a few modifications to the model to better address the data quality and availability of provincial SRBs in Pakistan. Subnational SRB models have been applied to other culturally and demographically heterogeneous countries with son preference such as Nepal (Chao, *et al.*, 2022) and Vietnam (Chao *et al.*, 2021c).

The outcome of interest $\Theta_{p,t}$, the SRB in Pakistan province p in year t is modeled as follows:

$$\Theta_{p,t} = b\Phi_{p,t} + \delta_p \alpha_{p,t},$$

b = 1.056 is the SRB baseline level for the entire Pakistan. The Pakistan SRB baseline b is estimated based on national SRB observations in Pakistan before the reference year 1970 (Chao *et al.*, 2019a, b). $p \in \{1,\ldots,k\}$ is the province index where k = 7. $t \in \{0,\ldots,h\}$ is the time index where t = 0 refers to the year 1980 and $t = h$ refers to the year 2050.

$\Phi_{p,t}$ follows an AR (1) time series model on the log scale, which captures the natural fluctuations of SRB in each province over time. The values of ρ and σ_ϵ (ρ = 0.9) and σ_ϵ = 0.004) were not estimated but were borrowed from a previous study (Chao *et al.*, 2019a,b), which robustly estimated the parameters from an extensive national SRB database. We assume that log ($\Phi_{p,t}$) is causal and weakly stationary AR (1) process with parameters ρ and σ_ϵ, and hence, log ($\Phi_{p,t-s}$) is uncorrelated with $\epsilon_{p,t}$ for all s>0. The unconditional variance at t = 0 is expressed as $\sigma_\epsilon^2 / \left(1-\rho^2\right)$ for log ($\Phi_{p,t}$). $\Phi_{p,t}$ is modeled as follows:

$$\log\left(\Phi_{p,t}\right) \sim \mathcal{N}\left(0, \sigma_\epsilon^2 / \left(1-\rho^2\right)\right), if\ t = 0,$$

$$\log\left(\Phi_{p,t}\right) = \rho \log\left(\Phi_{p,t-1}\right) + \epsilon_{p,t}, if\ t \in \{1,\cdots,h\},$$

$$\epsilon_{p,t} \sim_{i.i.d.} \mathcal{N}\left(0, \sigma_\epsilon^2\right).$$

δ_p is the binary identifier of the sex ratio transition at the provincial level with only two possible values 1 and 0. δ_p = 1 indicates an SRB imbalance in province p, whereas δ_p = 0 indicates no imbalance in province p. The provincial sex ratio transition identifier parameter δ_p is meant to detect whether the transition process exists based on the levels and trends in the SRB observations. To ensure that the probability parameter π_p lies in the interval [0, 1], we use the logit transformed π_p follows a hierarchical normal distribution with a global mean and variance μ_π and σ_π^2, respectively. δ_p follows a Bernoulli distribution:

$$\delta_p \mid \pi_p \sim \mathfrak{B}(\pi_p), for\ \ p \in \{1,\cdots,k\},$$

$$logit\ (\pi_p) \mid \mu_\pi, \sigma_\pi \sim \mathcal{N}\left(\mu_\pi, \sigma_\pi^2\right), for\ p \in \{1,\cdots,k\}.$$

Vague priors are assigned to the parameters related to the indicator that detects sex ratio transitions:

$$inverse - logit\left(\mu_\pi\right) \sim \mathcal{U}(0,1),$$

$$\sigma_\pi \sim \mathcal{U}(0,2).$$

$\alpha_{p,t}$ refers to the province-specific SRB imbalance process. The process is assumed to be non-negative and is modeled by a trapezoidal function representing the three consecutive stages (increase, stagnation, and decrease) of the sex ratio transition. The trapezoidal function specification of $\alpha_{p,t}$ is motivated by the patterns of national-level SRB observations in countries with strong statistical son preference to capture the three-stage transition process (Chao *et al.*, 2021a). The trapezoidal functional form for $\alpha_{p,t}$ can capture the shape of the observed SRB transition process according to Chao *et al.* (2021c) for those countries. $\alpha_{p,t}$ is modeled as:

$$\alpha_{p,t} = \left(\xi_p / \lambda_{1p}\right)\left(t - t_{0p}\right), if\ t_{0p} < t < t_{1p}$$

$$\alpha_{p,t} = \xi_p, if\ t_{1p} < t < t_{2p}$$

$$\alpha_{p,t} = \xi_p - \left(\xi_p / \lambda_{3p}\right)\left(t - t_{2p}\right), if\ t_{2p} < t < t_{3p}$$

$$\alpha_{p,t} = 0, if\ t < t_{0p}\ or\ t > t_{3p}$$

Where,

$$t_{1p} = t_{0p} + \lambda_{1p},$$

$$t_{2p} = t_{1p} + \lambda_{2p},$$

$$t_{3p} = t_{2p} + \lambda_{3p}.$$

The start year of the SRB inflation t_{0p} is modeled by a continuous uniform prior distribution with a lower bound at the year 1980 and an upper bound at the year 2050, respectively. For $p \in \{1,\ldots,k\}$, we have:

$$t_{0p} \sim \mathcal{U}(0,h).$$

The province-specific period lengths of the three stages of the SRB inflation (λ_{1p}, and λ_{3p}) are assigned with informative priors. The means of prior distributions are taken from a systematic study (Chao *et al.*, 2021a)

which modeled the sex ratio transition of multiple countries, including Pakistan. The standard deviations of prior distribution are set such that the CV is 0.1. The informative priors assist the provincial-level modeling of the sex ratio transition in Pakistan by exploiting the corresponding information at the national level. For $p \in \{1,\ldots,k\}$, we have:

$$\xi_p \sim \mathcal{N}_{(0,\infty)}\left(0.06, 0.006^2\right),$$

$$\lambda_{1p} \sim \mathcal{N}_{(0,\infty)}\left(11.0, 1.1^2\right),$$

$$\lambda_{2p} \sim \mathcal{N}_{(0,\infty)}\left(7.6, 0.8^2\right),$$

$$\lambda_{3p} \sim \mathcal{N}_{(0,\infty)}\left(16.1, 1.6^2\right).$$

2.2.2. Data quality model

r_i is the i[th] observed SRB in province p[i] in year t[i], where i indexes all SRB observations across the provinces over time. r_i is assumed to follow a normal distribution on the log scale with mean of $\log\left(\Theta_{p[i],t[i]}\right)$ and variance of σ_i^2:

$$\log\left(r_i\right)|\Theta_{p[i],t[i]} \sim \mathcal{N}\left(\log\left(\Theta_{p[i],t[i]}\right), \sigma_i^2 + \omega^2\right),$$
$$for\, i \in \{1,\cdots,n\},$$

Where, $n = 531$ is the total number of observations. σ_i^2 is the sampling error variance of log (r_i), which reflects the uncertainty in log-scaled SRB observations because of the survey sampling design. σ_i^2 is calculated using a jackknife method (Appendix A.1). ω^2 is the non-sampling error variance representing the uncertainty contributed by non-responses, recall errors, and data input errors. We assume that ω^2 is immeasurable and is estimated using the model by assigning a vague prior:

$$\omega \sim \mathcal{U}\left(0, 0.5\right).$$

2.2.3. Posterior distribution

Likelihood

For the i^{th} observation r_i, let $v_i = \log(r_i)$ and $V_{p,t} = \log\left(\Theta_{p,t}\right)$.

The likelihood on log-scale up to proportion is:

$$p\left(v_i|V_{p[i],t[i]},\omega\right) \propto \frac{1}{\sqrt{\sigma_i^2+\omega^2}} \exp\left\{-\frac{\left(v_i - V_{p[i],t[i]}\right)^2}{2\left(\sigma_i^2+\omega^2\right)}\right\}.$$

For $\in\{1,\ldots n\}$, the likelihood can be written as:

$$p\left(v_i|\Phi_{p[i],t[i]},\omega,\delta_{p[i]},\alpha_{p[i],t[i]}\right) \propto \frac{1}{\sqrt{\sigma_i^2+\omega^2}}$$
$$\exp\left\{-\frac{\left(v_i - b\Phi_{p[i],t[i]} - \delta_{p[i]}\alpha_{p[i],t[i]}\right)^2}{2\left(\sigma_i^2+\omega^2\right)}\right\}.$$

Posterior Density

The posterior density for $V_{p,t}$, the true SRB on the log scale for province p at time t, up to proportion is:

$$p\left(\begin{matrix}V_{1:k,0:h},\omega,\Phi_{1:k,0:h},\delta_{1:k},t_{0,1:k},\xi_{1:k},\lambda_{1,1:k},\lambda_{2,1:k},\lambda_{3,1:k}\\,\rho,\sigma_\epsilon,\mu_\pi,\sigma_\pi|v_{1:n}\end{matrix}\right) \propto$$
$$\frac{\left(1-\rho^2\right)^{k(h-1)/2}\prod_{\delta_p\in\{0,1\}}\pi_p^{\sum_{p=1}^k\delta_p}\left(1-\pi_p\right)^{k-\sum_{p=1}^k\delta_p}}{\sigma_\delta^{k(h-1)}\sigma_\pi^k\left[\prod_{t=1}^h\left(1-\rho^{2t}\right)^{k/2}\right]\prod_{i=1}^n\left(\sigma_i^2+\omega^2\right)^{1/2}}\times$$

$$\exp\left\{\sum_{p=1}^k\frac{2\mu_\pi\pi_p-\pi_p^2}{2\sigma_\pi^2}-\sum_{i=1}^n\frac{\left(v_i-b\Phi_{p[i],t[i]}-\delta_{p[i]}\alpha_{p[i],t[i]}\right)^2}{2\left(\sigma_i^2+\omega^2\right)}\right\}\times$$

$$\exp\left\{-\sum_{p=1}^k\sum_{t=1}^h\frac{\left(1-\rho^2\right)\left(\Phi_{p,t}-\rho^t\Phi_{p,0}\right)^2}{2\sigma_\epsilon^2\left(1-\rho^{2t}\right)}-\sum_{p=1}^k\frac{\sigma_\epsilon^2\Phi_{p,0}^2}{2\left(1-\rho^2\right)}\right\}.$$

2.2.4. Statistical computing and Bayesian Inference

We obtained posterior samples of all the model parameters and hyperparameters using a Markov chain Monte Carlo (MCMC) algorithm, implemented in the open source software R 4.2.1 (R Core Team, 2022) and JAGS 4.3.0 (Plummer, 2003), using R-packages R2jags (Su & Yajima, 2015) and rjags (Plummer, 2018). Results were obtained from 10 chains with a total of 5000 iterations in each chain, while the first 1000 iterations were discarded as burn-in. After discarding burn-in iterations and proper thinning, the final posterior sample size for each parameter by combining all chains is 25,000. The convergence of the MCMC algorithm and the sufficiency of the number of samples obtained were checked through visual inspection of trace plots and convergence diagnostics of Gelman & Rubin (1992), implemented in the coda R-package (Plummer *et al.*, 2006).

2.2.5. Post-modeling process

2.2.5.1. Identifying provinces of Pakistan with SRB imbalance

SRB imbalance in a Pakistan province is detected if $\delta_p = 1$ for more than 95% of the posterior samples (indicating SRB inflation).

2.2.5.2. Simulating SRB imbalance after 2020

In provinces of Pakistan without past/ongoing SRB inflation (assessed in the model), we simulate the SRB imbalance process after 2020 for different starting years of SRB inflation.

The simulated province-specific SRB imbalance process $\delta_p \alpha_{p,t}$ is based on posterior samples in the model. The simulated $\delta_p \alpha_{p,t}$ is added to the projected $\Theta_{p,t}$ for different starting years of SRB inflation in each province. The simulation process is detailed in the appendix.

Figure 2 shows the simulated SRB imbalance process $\delta_p \alpha_{p,t}$ in a Pakistan province, with a given start year of the inflation process t_0. The SRB inflation process spans 38 years. After approximately one decade, the imbalance reaches its maximum level and remains around that level for approximately 7 years. The SRB imbalance then deflated toward the normal/reference level of SRB (i.e., the SRB inflation becomes zero) over the next 15 years.

2.2.5.3. Computing the number of missing female births

Let $\psi_{p,t}$ and $\Psi_{p,t}^{inflation-free}$ denote the estimated and expected inflation-free numbers of female live births respectively, in province p in year t. The estimated and expected numbers of female birth are calculated as $\psi_{p,t} = B_{p,t}/(1+\Theta_{p,t})$ and $\Psi_{p,t}^{(inflation-free)} = \left(B_{p,t} - \Psi_{p,t}\right) / \Theta_{p,t}^{(inflation-free)}$, respectively.

The total number of births $B_{p,t}$ in a given province and year is obtained from Wazir and Goujon (2019). The number of inflation-free female births $\Psi_{p,t}^{(inflation-free)}$ is obtained from the estimated number of male births $(B_{p,t} - \psi_{p,t})$ and the inflation-free SRB $\Theta_{p,t}^{(inflation-free)} = b\Phi_{p,t}$ in the respective given province year. The number of missing female births is calculated using a method introduced in Dréze and Sen (1990), which was reviewed and validated in Guilmoto *et al.* (2020).

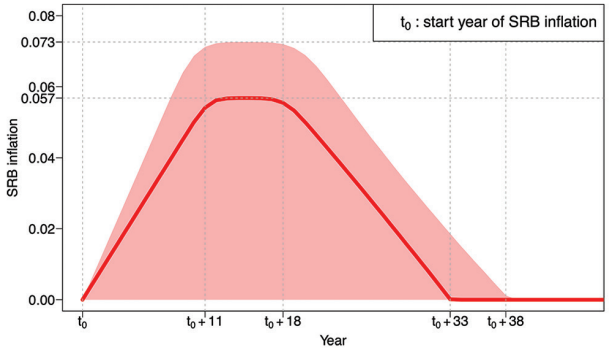

Figure 2. SRB inflation simulation of a Pakistan province
Note: Medians are in red curve. About 95% credible intervals are in red shades. t0 is the start year of the SRB inflation process.

The annual number of missing female births (AMFBs) in province p in year t is defined as: $\Psi_{p,t}^{(missing)} = \Psi_{p,t}^{(inflation-free)} - \Psi_{p,t}$. The cumulative number of missing female births (CMFB) from t_1 to t_2 in province p is obtained by adding the AMFB from year t_1 to year t_2: $\Lambda_{p,[t_1,t_2]}^{(missing)} = \sum_{t=t_1}^{t_2} \Psi_{p,t}^{(missing)}$.

3. Results

3.1. Levels, trends, and geographic disparities in provincial SRB estimates

Figure 3 shows an overview of the levels and trend in provincial SRBs in Pakistan from 1980 to 2020. The median estimates fluctuate around the national SRB reference level (1.056) except in Balochistan and Gilgit Baltistan. In Gilgit Baltistan, the SRB gradually increased from 1.058 (95% credible interval [1.041; 1.102]) in 1980 to 1.070 [1.047; 1.125] in 2016. After reaching its provincial maximum, the SRB continuously declines. The SRB in Gilgit Baltistan is not statistically significantly different from the national baseline level because the 95% credible intervals overlap with the national baseline throughout the whole period. However, the SRB in Balochistan is an outlier from the SRBs in all other provinces (Section 2.2.2 for details). The SRB estimates from 1980 to 2020, including uncertainty for the seven Pakistan provinces, are presented in Supplementary File 2 (https://doi.org/10.6084/m9.figshare.21548103).

Figure 4 illustrates the disparity in SRB across geographic locations. The SRB is most inflated in the southwest and northeast regions, including the Balochistan and Gilgit Baltistan provinces.

3.2. SRB imbalance at the provincial level

Table 2 lists the modeled SRB inflation probability in each Pakistan province. The probability of having a past or

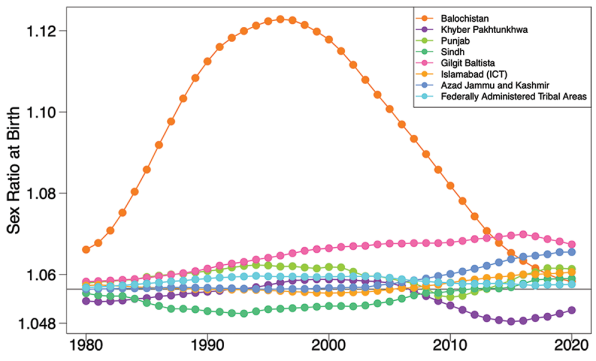

Figure 3. Median SRB estimates in Pakistan provinces over the 1980 – 2020 period

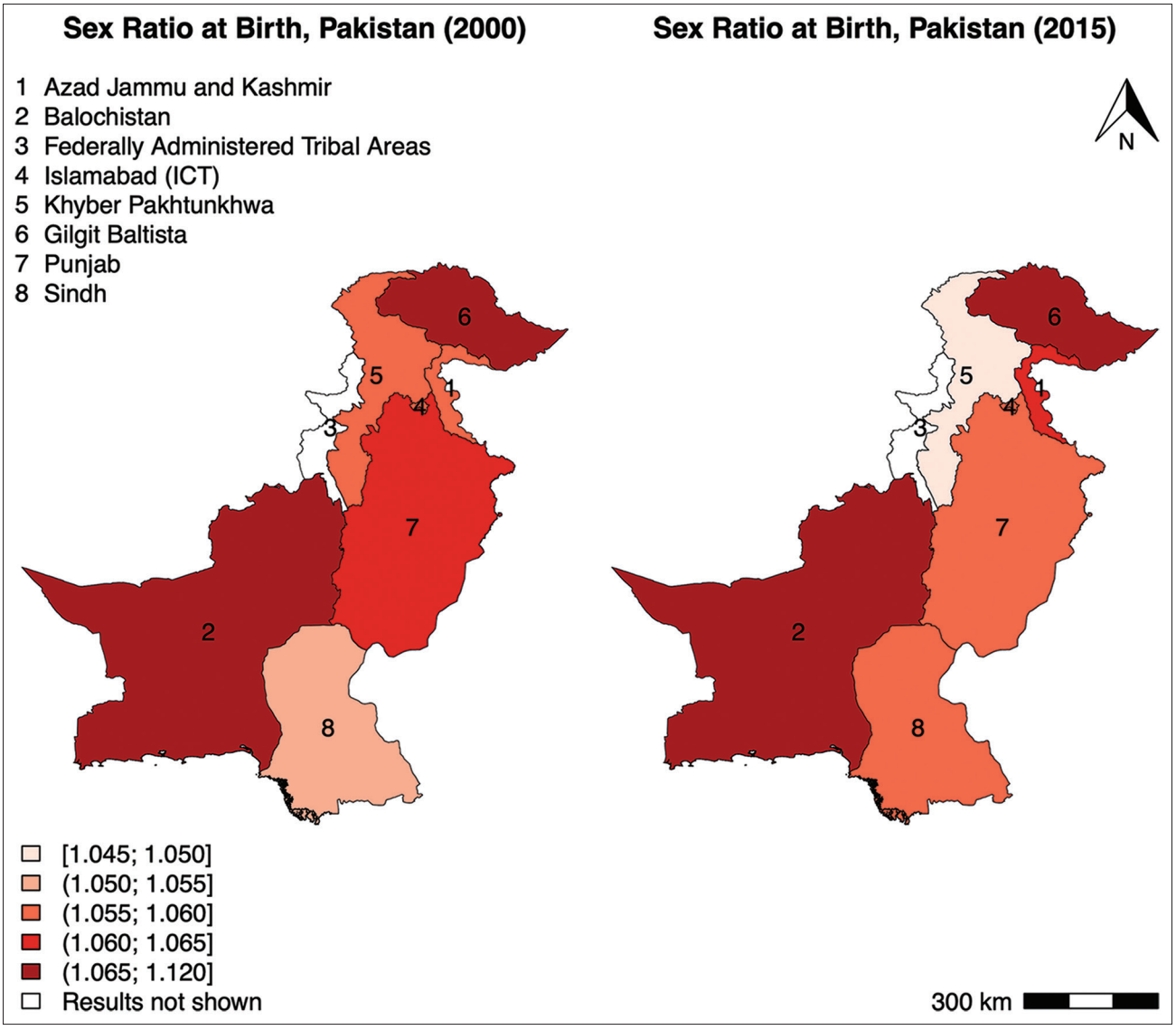

Figure 4. Geographic disparities of SRB estimates, 2000 and 2015
Note: Median estimates are presented. Results for Federally Administered Tribal Areas are omitted.

Table 2. SRB inflation probability by Pakistan province

Pakistan province	SRB inflation probability
Balochistan	100%
Gilgit Baltistan	65.4%
Azad Jammu and Kashmir	39.4%
Islamabad (ICT)	25.5%
Punjab	17.2%
Sindh	13.7%
Khyber Pakhtunkhwa	5.6%

Note: Provinces are listed in descending order of the inflation probability.
SRB: Sex ratio at birth.

ongoing SRB inflation is the highest in Balochistan at 100%. As the probability in Balochistan is the only probability above the cutoff value (95%), we identify Balochistan as the only province in Pakistan with SRB imbalance that happened before 2020.

Figure 5 illustrates the SRB model results in Balochistan. The SRB imbalance process in this province started in 1980 and ended around 2015. The maximum SRB in this province is estimated to occur in 1996, with SRB median estimate at 1.123 and the 95% credible interval at [1.100; 1.142]. The SRB in Balochistan is significantly above the national baseline (1.056) from 1986 to 2010. Over this

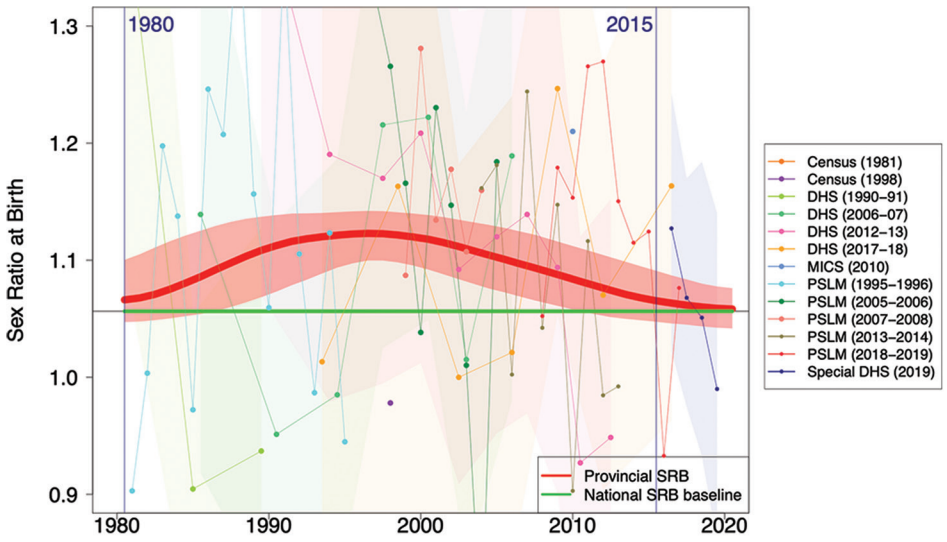

Figure 5. SRB estimates in Balochistan, 1980 – 2020

Note: Red line and shaded areas are the medians and 95% credible intervals of the province-specific SRB, respectively. The green horizontal line is the SRB baseline for the whole of Pakistan at 1.056 (Chao *et al*., 2019a). The SRB observations (dots) from the same source are connected with line segments of the same color. Shaded areas around the observation series represent the sampling errors in the series (quantified by twice the sampling standard errors). Blue vertical lines denote the start and end years of the sex ratio transition.

Table 3. Missing female births model results in Balochistan

Values are in thousands	Time period				
	1980 – 1990	1991 – 2000	2001 – 2010	2011 – 2020	1980 – 2020
Estimated female births	124.2	124.2	153.2	174.9	143.6
	[122.3; 125.8]	[123.3; 125.5]	[151.5; 155.1]	[173.1; 176.2]	[142.9; 144.4]
Expected female births	127.8	131.7	159.2	176.4	148.3
	[126.3; 129.6]	[130.5; 132.7]	[157.4; 160.9]	[175.2; 178.2]	[147.6; 149.0]
AMFB	3.7	7.6	6.0	1.8	4.7
	[0.7; 7.4]	[5.0; 9.4]	[2.3; 9.4]	[0.1; 5.1]	[3.4; 6.1]
CMFB	40.2	76.1	60.1	18.2	194.6
	[7.4; 81.2]	[50.0; 93.9]	[23.5; 94.1]	[0.5; 51.0]	[140.0; 248.9]

Note: All values are in thousands. Numbers in front of the brackets are the posterior medians, and those inside brackets are the 95% credible intervals. The estimated female births, expected female births, and AMFBs are averaged over each period. The CMFBs are the cumulative values over each period. AMFB: Annual number of missing female births. CMFB: Cumulative number of missing female births.

period, the lower bound of the 95% credible intervals of SRB exceeded the national baseline.

3.3. Missing female births before 2020 in Balochistan

Table 3 lists the missing female births in Balochistan over different time periods. From the estimated and expected female births, we demonstrate the baseline magnitude of female births in Balochistan. The estimated average AMFB over the four decades from 1980 to 2020 is 4.7 [3.4; 6.1] thousand. On a decade-by-decade basis, the average AMFB increased from 3.7 [0.7; 7.4] in 1980 – 1990 to 7.6 [5.0; 9.4] in 1991 – 2000, then gradually declined from 2000, reaching 1.8 [0.1; 5.1] in the 2011 – 2020 period. Consequently, as the cumulative result of the AMFB, the CMFB between 1980 and 2020 is estimated at 194.6 [140.0; 248.9] in Balochistan province. Our results are consistent with previous results (Qayyum & Rehan, 2017) of sex-selective abortion in Balochistan. Although the data of that study were collected only from the rural areas (and hence may not be provincially representative), Balochistan had the highest rate of sex-selective abortion during the 2011 – 2014 period.

3.4. Scenario-based missing female births simulation after 2020

Although we identify Balochistan as the only province with past/ongoing SRB inflation, we do not rule out the possibility that the imbalanced SRB will emerge in other

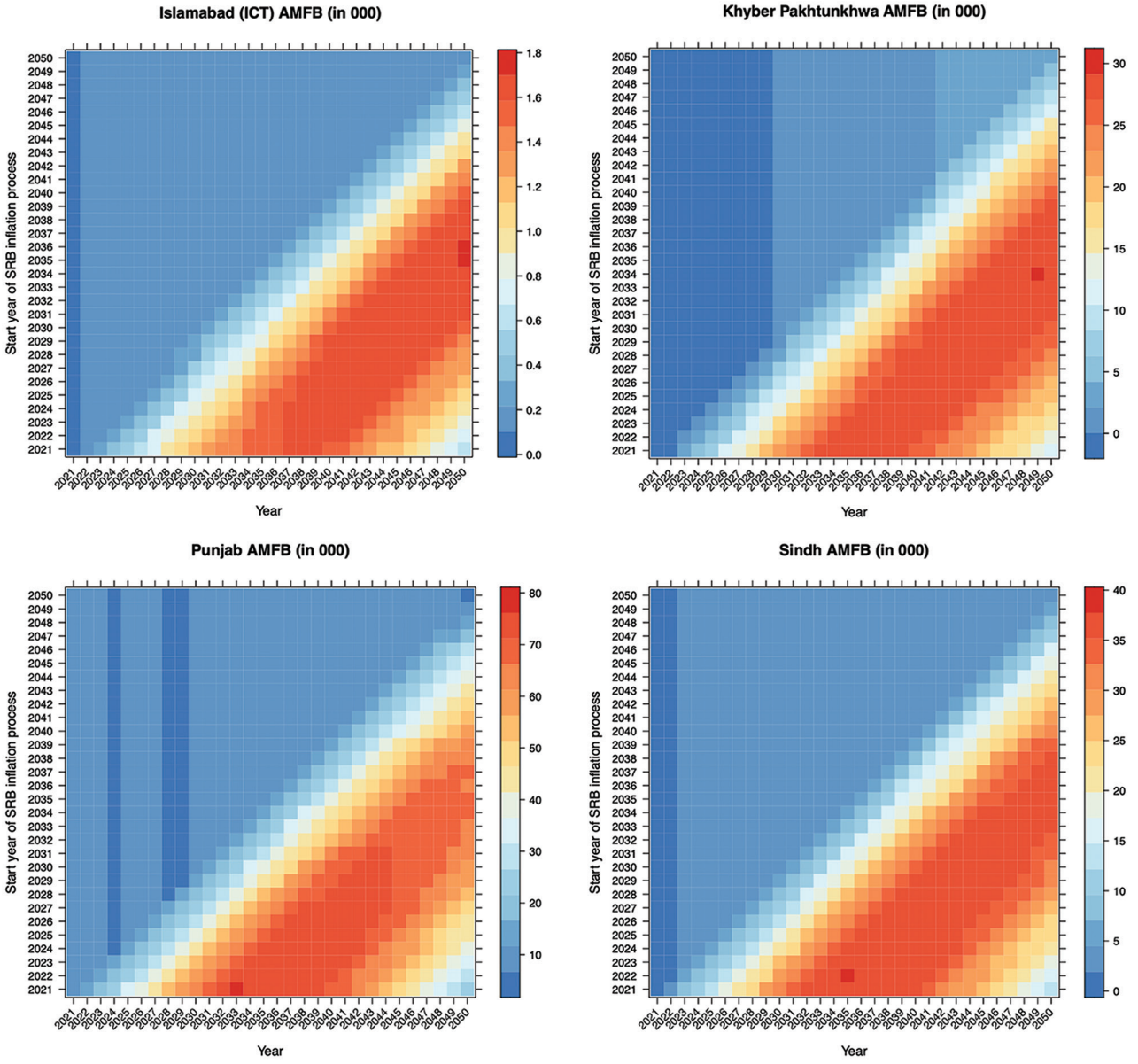

Figure 6. Post-2020 missing female births simulation in provinces without past imbalanced SRB

Note: Median projections of AMFB (in thousand) are presented. AMFB: Annual number of missing female births. Results are shown for provinces with projections of annual total births. X-axis: Projection period 2021 – 2050. Y-axis: Simulation scenarios with different start years of SRB inflation from 2021 to 2050.

provinces. Hence, we present the results of scenario-based projections of missing female births in provinces without current SRB inflation.

Figure 6 shows the AMFB results in four provinces: Islamabad (ICT), Khyber Pakhtunkhwa, Punjab, and Sindh, in which the projected annual total numbers of births are available and are not identified with past SRB inflation. Each row in each heatmap shows the simulated AMFBs over the projection period 2021 – 2050 under the scenario that the SRB inflation process starts in a certain year.

In general, the later the assumed start year of the SRB imbalance process, the AMFBs in more years are projected near zero. In Figure 6, the heatmap, these trends manifested as increasing extents of blue areas as the rows are traversed from bottom to top. In the heatmap, darker blue means a smaller number of missing female births while redder means a higher number of missing female births. This implies that while the start year of the SRB inflation process delays year by year (i.e., moving up on the y-axis), there are delayed effects on the occurrence of

the number of missing female births. When the simulated SRB becomes imbalanced during the projection period, the three stages of the sex ratio transition (increase, stagnation, and return to normal) are visible in the resulting AMFBs. However, the AMFB is influenced not only by the SRB imbalance process but also by the levels and trends in the total number of births over time. Accounting for both the SRB inflation and the total number of births, we project the maximum AMFB for different combinations of start-year scenarios and the year in the projection period. In Islamabad (ICT), the maximum AMFB is projected to occur in 2050 when the SRB imbalance process starts in 2035, with the AMFB at 1.7 thousand. In Khyber Pakhtunkhwa, the maximum AMFB is projected at 29.2 thousand in 2049 when the SRB inflation starts in 2034. The largest AMFB in Punjab is projected to occur in 2033 when the SRB inflation stats in 2021 and its projected value is 76.2 thousand. In Sindh, when the SRB imbalance process starts in 2022, the maximum AMFB across all scenarios is projected to occur in 2035 at 37.8 thousand.

4. Discussion

Policy planners can prepare guidelines for preventing prenatal sex discrimination using scenario-based projections of the number of missing female births in the provinces without ongoing SRB inflation. A recent study provided missing female birth projections at the national level for all countries in the world (Chao *et al.*, 2021b). According to that study, if sex-selective abortion were to happen in Pakistan national wide, the missing female births may contribute as high as 14% of the global numbers during the 2021 – 2100 period. Our study reveals the potential future missing female births in Pakistan in four provinces, namely, Islamabad (ICT), Khyber Pakhtunkhwa, Punjab, and Sindh. For different start years of the SRB imbalance in each province, we identify the years in which the number of missing female births will possibly deficit the greatest. The projections results reflect the fact that the number of missing female births is a combined effect of the SRB inflation process and fertility transition. Given the speed of the fertility transition in Pakistan, and the estimates from the 2017 – 2018 DHS, we revealed that the decline in fertility rates in Pakistan has slowed at both the national and subnational levels since the 1990s, and the total fertility rate at 3.6 remains higher than in neighboring countries (National Institute of Population Studies (NIPS) [Pakistan] & ICF, 2019).

The findings of this study reinforce the persistence of strong son preferences in Pakistan and identified high-level distortion in Balochistan. The three conditions of Guilmoto (2009) hypothesis for the distortion of SRB in Pakistan are currently well aligned: Inherited gender discrimination, preference for large families, and technological advancement. Pakistan fares poorly on gender equality, ranking 135 of 191 countries on the Gender Inequality Index (United Nations Development Programme, 2022). The deep-rooted social and cultural norms and practices continue to be the underlying cause of gender inequalities.

Furthermore, inflated SRB influences the demand of a larger number of children, currently manifesting as higher fertility in Pakistan for the sake of more sons. The fertility stagnation in Pakistan since the onset of the 21st century is evident primarily attributed to the higher ideal family size with at least two sons (Wazir, 2018). The imbalanced SRB leads to prolonged consequences in both demographic and social aspects. Imbalanced SRB is one of the main factors that lead to the phenomenon of "missing women" first endeavored by Amartya Sen, referring to the females that should have survived or been born in the absence of sex discrimination and excess mortality among females (Sen, 1990). A large number of "missing women" results in a marriage squeeze, increased levels of antisocial behavior and violence, and may eventually have a long-term impact on stability and social development.

Despite the strong son preference persists in Pakistan, one study implied that son preference has not resulted in nationwide sex-selective abortions but occurred in subpopulations such as in urban clinics (Sathar *et al.*, 2015). Although abortion is illegal in Pakistan, the abortion rate significantly increased from 27 to 50/1000 for women aged 15 – 49 over the period 2000 – 2012. Meanwhile, Balochistan experienced the highest abortion rate of 60/1000 for women aged 15 – 49 among the provinces (Sathar *et al.*, 2014). In addition, our study also reinforces the distortion of SRB in Balochistan is highest among the provinces. These numbers are associated with well-documented demographic phenomena of "missing women" at the national and provincial levels in Pakistan. Although, the compelling evidence of sex selection and excess mortality are not prevailing in Pakistan on the national level, the absence of evidence is not the evidence of absence. Our subnational study pinpoints the disparity in SRB and sex-selective abortion on the provincial level that is masked by national-level results. There is an urgent need to generate high-quality data in Pakistan, particularly through census at the subnational level followed by in-depth research on the persistence of discriminatory practices of sex selection and excess mortality for females.

This is the first study on estimating SRB in Pakistan from 1980 to 2020 and provides scenario-based projections of missing female births up to 2050 by province based on a Bayesian hierarchical time series model. Our results revealed important SRB disparity across geographic locations in Pakistan. Among the seven provinces included in the study,

Balochistan presents a decisively imbalanced SRB. In the other provinces without the existing SRB inflation, we demonstrate important disparities in the occurrences and quantities of female birth deficits before 2050.

5. Conclusions

Our study provides model-based and data-driven SRB estimates and projections for provinces in Pakistan from 1980 to 2050. Our model results demonstrate important disparities in SRB levels and trends across provinces over time. Balochistan is identified as the only province in Pakistan with an existing SRB imbalance and, consequently missing female births. In future work, in-depth provincial studies and the collection of high-quality birth data are required to monitor subnational SRB disparities in Pakistan.

Effective program and policy solutions to curb sex discrimination remain elusive in Pakistan because the practices, leading to excess mortality among females and sex selection, are often poorly understood. Therefore, the institutional response is primarily focused on the improvement of the provision of health care. The last two decades have witnessed the adoption of several pro-women laws such as prevention of sexual violence and harassment, protection, domestic violence, and early marriages. However, the implementation remains challenging, primarily because of federal and provincial autonomy to deliver basic social services. Advancing gender equality and discriminatory practices require accountability mechanisms for policy implementation and enforcement of laws, adequate financing at the provincial level, and community engagement to address discriminatory gender and social norms.

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Conflict of interest

No conflicts of interest were reported by all authors.

Author contributions

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Formal analysis: Fengqing Chao and Hernando Ombao
Supervision: Muhammad Asif Wazir
Validation: Fengqing Chao and Hernando Ombao
Writing – original draft: Fengqing Chao
Writing – review & editing: Fengqing Chao, Muhammad Asif Wazir, and Hernando Ombao

Ethics approval and consent to participate

The human data used in our study are secondary and publicly available datasets from surveys and censuses. The survey data are available at https://dhsprogram.com/ for the DHS Program and at https://mics.unicef.org/surveys for MICS. The census data are available from IPUMS International at https://international.ipums.org/international/.

Consent for publication

Not applicable.

Availability of data

Supplementary File 1: Pakistan provincial SRB database. DOI: https://doi.org/10.6084/m9.figshare.21548082

Supplementary File 2: SRB estimates by Pakistan province from 1980 to 2020. DOI: https://doi.org/10.6084/m9.figshare.21548103

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Appendix

Data processing

Appendix A. Sampling errors in the DHS and MICS data

Both Demographic and Health Survey (DHS) and Multiple Indicator Cluster Survey (MICS) provide individual-level data with the full birth history of each woman of reproductive age interviewed during the survey fieldwork period. We calculated the sampling error in the log-transformed sex ratio at birth (SRB) obtained from the DHS and MICS data series using the jackknife method (Efron & Gong, 1983; Efron & Tibshirani, 1994; ICF International, 2012). For a certain DHS or MICS data series, let U denote the total number of clusters (based on the cluster/primary sampling unit numbers in the survey data (Verma & Le 1996)). The u-th partial prediction of SRB is determined by the following equation:

$$r_{-u} = \frac{\sum_{n=1}^{N} I_n\left(x_n = male; d_n \neq u\right) w_n}{\sum_{n=1}^{N} I_n\left(x_n = female; d_n \neq u\right) w_n}, for\, u \in \{1,...,U\},$$

Where, n indexes the live births in each state-survey-year; N is the total number of live births; and x_n, d_n, and w_n are the sex, cluster number, and sampling weight for the n-th live birth, respectively. The sampling weight of each birth w_n is extractable from the survey data and reflects the survey sampling design (Verma & Le, 1996). We define I_n (.) = 1 if the condition inside the brackets is true and I_n (.) = 0 otherwise. The u-th pseudo-value estimate of the SRB on the log-scale is:

$$\log\left(r\right)_u^* = U\log\left(r^{'}\right) - \left(U-1\right)\log\left(r_{-u}\right), where$$

$$r^{'} = \frac{\sum_{n=1}^{N} I_n\left(x_n = male\right) w_n}{\sum_{n=1}^{N} I_n\left(x_n = female\right) w_n}.$$

The sampling variance is:

$$\sigma^2 = \frac{\sum_{u=1}^{U}\left(\log\left(r\right)_u^* - \overline{\log\left(r\right)_u^*}\right)^2}{U\left(U-1\right)}, where$$

$$\overline{\log\left(r\right)_u^*} = \frac{1}{U}\sum_{u=1}^{U}\log\left(r\right)_u^*.$$

In the DHS or MICS data, the annual log-transformed SRB observations are merged such that the coefficient of variation (CV) for log-transformed SRB is below 0.1 or the merged period reaches 5 years (Pedersen & Liu, 2012).

For a certain DHS/MICS data series, let $\{t_n, t_{n-1},\ldots,t_1\}$ be years with recorded births from recent to past. The merge starts from the most recent year t_n and goes back year by year to $t_{n-1},\ldots,1$. The process is performed by the following algorithm for each DHS and MICS data series:

Step #	Merging process of DHS and MICS data
1	for $t \in \{t_n, t_n-1,\ldots, t_1\}$ do
2	if $t=t_n$ then
3	Compute σ as explained above. Compute
4	if CV<0.1 or $t_n - t_{n-1} > 1$ then
5	stop and move to the previous time point
6	else
7	Set $t=t_{n-1}$, merge births from tn and, t_{n-1} by summing them up
8	Repeat step 3
9	if CV<0.1 or $t_{n-1} - t_{n2} >1$ or $t_n - t_{n-1} = 5$ then
10	stop and move to the previous time point
11	else
12	Set $t=t_{n-2}$, merge births from t_n, t_{n-1}, t_{n-2} by summing them up
13	Repeat steps 8 – 12 for $t \in \{t$n−2,…, $t1\}$

A. 1. Further explanations of the motivation and assumptions of merging DHS/MICS data

The above algorithm is to merging observations from single calendar years into observations over short time periods from DHS and MICS surveys where full birth histories are available. The merge refers to summing up the number of sex-specific births across multiple years before computing the SRB for that period. The purpose of the merging process is meant to reduce uncertainties associated with observations to a reasonable level. Without merging the births from each calendar year, the sampling errors in these population indicators become unacceptably large due to the small sample sizes (Pedersen & Liu, 2012). The underlying assumptions for the following expressions are:

- Step 1 for $t \in \{t_n, t_{n-1},\ldots,t_1\}$ do: this means that we are merging the 1-year observations from the most recent year t_n to the earliest year with data t_1. The reason we merge the observations backward in time rather than merge forward is that: Usually in countries where DHS and MICS surveys are conducted, more births were sampled in recent years than in older years. This is largely due to the improvement of surveying technology, more mothers were still alive to recall recent births than births born decades ago, and less recall bias happened to births born in recent years than in earlier periods. Hence, the 1-year observations in recent years are usually less likely to be merged

compared to those in earlier periods because the sample sizes are usually larger in recent years. By merging backwards, we are able to preserve more observations in recent years without merging them. If we merge forward, it is likely that some of the observations in recent periods would be merged with observations in early periods.

- Step 4 if $CV < 0.1$ or $t_n - t_{n-1} > 1$: When time t is at the most recent year t_n, the condition to stop merging the birth to t_{n-1}, the previous year where data is available is (i) the $CV < 0.1$; or (ii) t_{n-1} is not the adjacent calendar year of t_n, i.e. $t_n - t_{n-1} > 1$.
- Step 9 if $CV < 0.1$ or $t_{n-1} - t_{n-2} > 1$ or $t_n - t_{n-1} = 5$: When time t is not at the most recent year t_n, the condition to stop merging the birth to t_{n-2} is (i) the $CV < 0.1$; or (ii) t_{n-2} is not the adjacent calendar year of t_{n-1}, that is, $t_{n-2} - t_{n-2} > 1$; or (iii) the merged period is already 5 years, that is, $t_n - t_{n-1} = 5$. We set the upper limit of merging period to 5 years because we want to still generate a time series of observations even after merging. Without setting an upper limit, too few numbers of observations may be generated just to satisfy the CV condition. The 5-year upper bound follows the conventional period length used in other population indicators as suggested by Pedersen & Liu, 2012.

Appendix B. Bayesian model for provincial SRB estimation and projection

B. 1. Notations

The notations are listed in the table below.

B. 2. Scenario-based simulated projections for SRB inflation

The province-specific SRB imbalance process $\delta_p \alpha_{p,t}$ is simulated using posterior samples from the model. The simulated $\delta_p \alpha_{p,t}$ is added to the projected $b\Phi_{p,t}$ for different starting years of the SRB inflation in each province.

For $g \in \{1,\ldots,G\}$, the g^{th} simulated SRB inflation is denoted as $\alpha_{p,t}^{'(g)}\delta_p^{(g)}$. $\delta_p^{(g)}$ is the g^{th} posterior sample of parameter δ_p. $\alpha_{p,t}^{'(g)}$ is the g^{th} simulated SRB imbalance process, with the start year of inflation fixed at $t_0 \in \{2021,...,2050\}$. $\alpha_{p,t}^{'(g)}$ is simulated as below for $g \in \{1,...,G\}$:

$$\alpha_{p,t}^{'(g)} = \left(\xi_p^{(g)} / \lambda_{1p}^{(g)}\right)\left(t - t_0\right), if\ t_0 < t < t_{1p}^{(g)}$$

$$\alpha_{p,t}^{'(g)} = \xi_p^{(g)}, if\ t_{1p}^{(g)} < t < t_{2p}^{(g)}$$

$$\alpha_{p,t}^{'(g)} = \xi_p^{(g)} - \left(\xi_p^{(g)} / \lambda_{3p}^{(g)}\right)\left(t - t_{2p}^{(g)}\right), if\ t_{2p}^{(g)} < t < t_{3p}^{(g)}$$

Symbol	Description
	Index
i	Indicator of the i^{th} SRB observation across all province-years, $i \in \{1,\ldots,531\}$
t	Indicator of year, $t \in \{1980,\ldots,2050\}$
p	Indicator of provinces of Pakistan, $p \in \{1,7\}$
Unknown parameters	
$\Theta_{p,t}$	Model fitting to the true SRB in Pakistan province p in year t
$\Phi_{p,t}$	Province year-specific multiplier for capturing the natural fluctuation in SRBs around the national baseline b in Pakistan province p in year t
$\alpha_{p,t}$	SRB imbalance in Pakistan province p in year t
t_{0p}	Start year of SRB inflation in Pakistan province p
δ_p	Indicator of the presence ($\delta_p = 1$) or absence ($\delta_p = 0$) of SRB inflation in Pakistan province p
ξ_p	Maximum level of SRB inflation in Pakistan province p
λ_{1p}	Period length of the increase stage of the sex ratio transition in Pakistan province p
λ_{2p}	Period length of the stagnation stage of the sex ratio transition in Pakistan province p
λ_{3p}	Period length of the decrease stage of the sex ratio transition in Pakistan province p, which returns the SRB to the national SRB baseline
ω	Non-sampling error
r_i	The i^{th} SRB observation
σ_i	Sampling error for the ith SRB observation (computed in Appendix A.1)
b	Baseline level of SRB over the whole of Pakistan (Chao *et al.*, 2019a), where $b = 1.063$
ρ	Autoregressive Indicator of $\Phi_{p,t}$, where $\rho = 0.9$ (Chao et al., 2019a, b)
σ_ϵ	Standard deviation of distortion parameter for $\Phi_{p,t}$, where $\sigma_\epsilon = 0.004$ (Chao et al., 2019a, b)

$$\alpha_{p,t}^{(g)} = 0, if\ t < t_0\ or\ t > t_{3p}^{(g)}$$

Where,

$t_{1p}^{(g)} = t_0 + \lambda_{1p}^{(g)}$, $t_{2p}^{(g)} = t_{1p}^{(g)} + \lambda_{2p}^{(g)}$, $t_{3p}^{(g)} = t_{2p}^{(g)} + \lambda_{3p}^{(g)}$.

$\xi_p^{(g)}$, $\lambda_{1p}^{(g)}$, $\lambda_{2p}^{(g)}$, and $\lambda_{3p}^{(g)}$ are the g[th] posterior samples of parameters ξ_p, λ_{1p}, λ_{2p}, and λ_{3p}.

Appendix C. Model validation

The performance of the inflation model was evaluated by two approaches: (1) Out-of-sample validation and (2) one-province simulation.

C. 1. Out-of-sample validation

We leave out 12% of the observations since the data collection year 2018 instead of reference year, which has been used for assessing model performance of demographic indicators largely based on survey data (Alkema *et al.*, 2012; Alkema *et al.*, 2014; Chao *et al.*, 2018a,b). There are 64 left-out observations from six Pakistan provinces. After leaving out the data, we fit the model to the training dataset and obtain point estimates and credible intervals that would have been constructed from the available dataset in the selected survey year. Based on the training dataset, we also generate the prediction distribution for each left-out observation.

We calculate the median errors and median absolute errors in the left-out observations. The errors are defined as $e_j = y_j - \widetilde{y_j}$, where $\widetilde{y_j}$ refers to the posterior median of the predictive distribution based on the training dataset for the j^{th} left-out observation y_j. The coverage is given by $1/J \sum_o I[y_j \geq l_j]\ I[y_j \leq u_j]$, where J refers to the number of left-out observations, and l_j and u_j correspond to the lower and upper bounds, respectively, of the 95% prediction interval of the j^{th} left-out observation y_j.

The validation measures are calculated for 1000 sets of left-out observations where each set contains one randomly selected left-out observation from each Pakistan province. The reported validation results are based on the mean outcomes of the 1000 sets of left-out observations. This technique of validation exercise is used to reduce the correlation of validation results within each province and has been used in validation exercises in the previous studies (Alkema *et al.*, 2014; Chao *et al.*, 2018a; You *et al.*, 2015).

Specifically, the final validation results regarding the left-out observations are calculated as follows:

1. For $k \in \{1,\ldots,1000\}$, we select a set of left-out observations $\{y_{k,1},\ldots y_{k,p},\ldots, y_{k,6}\}$. $y_{k,p}$ is the only selected left-out observation from province p and six provinces have left-out observations. Hence, we have $y_{k,1}$ to $y_{k,6}$.
2. For the k^{th} set of left-out observations $\{y_{k,1},\ldots y_{k,p},\ldots, y_{k,6}\}$, we can get the following results:
   - Corresponding errors $\{e_{k,1},\ldots e_{k,p},\ldots, e_{k,6}\}$ for these selected left-out observations.
   - Median of this set of error: medium $(e)_k$.
   - Coverage for this set: Coverage $e_k = \frac{1}{6}\sum_{p=1}^{6} I\left[y_{k,p} \geq l_{k,p}\right] I\left[y_{k,p} \leq u_{k,p}\right]$. Here $l_{k,p}$ and $u_{k,p}$ correspond to the lower and upper bounds of the 95% prediction interval of the left-out observation $y_{k,p}$.
3. Compute the mean of these results for the 1000 set of observations:
   - Corresponding errors $\{e_{k,1},\ldots e_{k,p},\ldots, e_{k,6}\}$ for these selected left-out observations.
   - Final median of error: $\frac{1}{1000}\sum_{k=1}^{1000} median(e)_k$.
   - Final coverage: $\frac{1}{1000}\sum_{k=1}^{1000} coverage_k$.

For the point estimates obtained from the full and training datasets, we define the errors in the true SRB as $e(\Theta)_{p,t} = \hat{\Theta}_{p,t} - \tilde{\Theta}_{p,t}$, where $\hat{\Theta}_{p,t}$ is the posterior median in province p in year t obtained from the full dataset, and $\tilde{\Theta}_{p,t}$ is the posterior median in the same province-year obtained from the training dataset.

Similarly, the error in the sex ratio transition process with probability is defined as $e(\alpha\delta)_{p,t} = \hat{\alpha}_{p,t}\hat{\delta}_p - \tilde{\alpha}_{p,t}\tilde{\delta}_p$. The coverage is computed similarly to the left-out observations and is based on the lower and upper bounds of the 95% credible interval of $\tilde{\Theta}_{p,t}$ from the training dataset.

C.2. One-province simulation

We assess the inflation model performance in a one-province simulation setting. We simulate SRB for a province prior observing data. In this simulation exercise, we consider all observations as the test data and simulate the SRB using the posterior samples of only the global parameters (instead of province-specific parameters) obtained from the sex ratio transition model using the full dataset. Hence, we simulate the SRB for a province without data and check how well the simulated results can align and cover the SRB observations in each province.

The g^{th} simulated SRB for a "new" province $\Theta(new)_t^{(g)}$ in year t are obtained as follows for $g \in \{1,\ldots,G\}$:

$$\Theta(new)_t^{(g)} = b\Phi(new)_t^{(g)} + \alpha(new)_t^{(g)}\delta(new)^{(g)}$$

Where the simulated $\Phi(new)_t^{(g)}$, $\alpha(new)_t^{(g)}$ and $\delta(new)^{(g)}$ refer to a "new" province without data. This simulation follows the model specifications of these parameters without considering any province-specific data. In particular, $\Phi(new)_t^{(g)}$ is simulated as:

$$\log\left(\Phi(new)_t^{(g)}\right) \sim \mathcal{N}\left(0, \frac{\left(\sigma_\epsilon^{(g)}\right)^2}{1-\left(\rho^{(g)}\right)^2}\right), if\ t = 1980,$$

$$\log\left(\Phi(new)_t^{(g)}\right) = \rho^{(g)}\log\left(\Phi(new)_{t-1}^{(g)}\right) + \epsilon_t^{(g)}, if\ t \in \{1981, \cdots, 2020\},$$

$$\epsilon_t^{(g)} \sim_{i.i.d.} \mathcal{N}\left(0, \left(\sigma_\epsilon^{(g)}\right)^2\right).$$

δ (new)$^{(g)}$ is simulated as:

$$logit\left(\pi^{(g)}\right) \sim \mathcal{N}\left(\mu_\pi^{(g)}, \left(\sigma_\pi^{(g)}\right)^2\right),\ \delta(new)^{(g)} \sim \mathcal{B}\left(\pi^{(g)}\right),$$

$\alpha(new)_t^{(g)}$ is simulated as:

$$\xi^{(g)} \sim \mathcal{N}_{(0,\infty)}\left(0.06,\ 0.006^2\right),\ \lambda_1^{(g)} \sim \mathcal{N}_{(0,\infty)}\left(11.0, 1.1^2\right),$$

$$\lambda_2^{(g)} \sim \mathcal{N}_{(0,\infty)}\left(7.6, 0.8^2\right),\ \lambda_3^{(g)} \sim \mathcal{N}_{(0,\inf)}\left(16.1,\ 1.6^2\right),$$

$$t_0^{(g)} \sim \mathcal{U}\left(1970,\ 2050\right),$$

$$\alpha(new)_t^{(g)} = \left(\xi^{(g)} / \lambda_1^{(g)}\right)\left(t - t_0^{(g)}\right), if\ t_0^{(g)} < t < t_1^{(g)},$$

$$\alpha(new)_t^{(g)} = \xi^{(g)}, if\ t_1^{(g)} < t < t_2^{(g)}$$

$$\alpha(new)_t^{(g)} = \xi^{(g)} - \left(\xi_p^{(g)} / \lambda_3^{(g)}\right)\left(t - t_2^{(g)}\right), if\ t_2^{(g)} < t < t_3^{(g)}$$

$$\alpha(new)_t^{(g)} = 0, if\ t < t_0^{(g)}\ or\ t > t_3^{(g)}$$

Where,

$$t_1^{(g)} = t_0^{(g)} + \lambda_1^{(g)},\ t_2^{(g)} = t_1^{(g)} + \lambda_2^{(g)},\ t_3^{(g)} = t_2^{(g)} + \lambda_3^{(g)}.$$

After generating the simulated values, we calculate results as described for the out-of-sample validation (Appendix C.1.1).

Appendix D. Validation and simulation results

Table D.1 summarizes the results of the left-out SRB observations in the out-of-sample validation exercise and one-province simulation. The median errors are nearly zero in the left-out observations. Although the median absolute errors are slightly higher than the median errors, the average coefficient of variance of the absolute errors for left-out observations (calculated as absolute errors divided by the left-out observation values) is only 5.6%. The coverage of the 95% and 80% prediction intervals is more conservative than expected. The wider-than-expected prediction interval in the left-out observations can be primarily attributed to larger uncertainty in more recent observations.

Table D.2 compares the model estimates obtained from the full dataset and the training set in the out-of-sample validation exercise. Here, we examined the model estimates of the true SRB $\Theta_{p,t}$ and the inflation process with province-specific probability $\delta_p \alpha_{p,t}$. The median errors and the median absolute errors are close to zero.

In summary, the validation results indicate reasonably good calibrations and prediction power of the inflation model with conservative credible intervals.

Table D.1. Validation and simulation results for left-out SRB observations

	Validation out of sample	Simulation
# Province in test dataset	6	8
Median error	0.020	0.003
Median absolute error	0.047	0.071
Below 95% prediction interval (%)	0.0	0.2
Above 95% prediction interval (%)	0.0	3.2
Expected (%)	2.5	2.5
Below 80% prediction interval (%)	0.0	7.6
Above 80% prediction interval (%)	8.0	9.2
Expected (%)	10	10

Note: Error is defined as the difference between a left-out SRB observation and the posterior median of its predictive distribution. SRB observations with data collection years since 2018 are left out. Numbers in the parentheses after the proportions indicate the average number of left-out observations that fall below or above their respective 95% and 80% prediction intervals.

Table D.2. Validation results of estimates based on the training set

Model validation	$\Theta_{p,t}$			$\delta_p \alpha_{p,t}$		
(Out-of-sample)	**1995**	**2005**	**2015**	**1995**	**2005**	**2015**
Median error	0.001	0.001	0.002	0.000	0.000	0.000
Median absolute error	0.001	0.001	0.004	0.000	0.000	0.000
Below 95% credible interval (%)	0.0	0.0	0.0	0.0	0.0	0.0
Above 95% credible interval (%)	0.0	0.0	0.0	0.0	0.0	0.0
Expected (%)	≤2.5	≤2.5	≤2.5	≤2.5	≤2.5	≤2.5
Below 80% credible interval (%)	0.0	0.0	0.0	0.0	0.0	0.0
Above 80% credible interval (%)	0.0	0.0	0.0	0.0	0.0	0.0
Expected (%)	≤10	≤10	≤10	≤10	≤10	≤10

Note: Error defines the differences between the model estimates (i.e., $\Theta_{p,t}$ or $\delta_p \alpha_{p,t}$) obtained from the full and training datasets, and proportions refer to the proportions (%) of countries in which the median estimates from the full dataset fall below or above their respective 95% and 80% credible intervals, respectively, in the training set.

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RESEARCH ARTICLE

Molecular characterization of beta-thalassemia reveals the presence of common mutations in the population of Himalayan region: Garhwal (Uttarakhand), India

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Abstract

Thalassemia is a dreadful heritable hemolytic disease, characterized by a genetic mutation in the hemoglobin subunit beta (HBB) gene. Mutation in HBB gene completely halts the production of the beta-globin protein, which leads to the defective production of functional hemoglobin. The prevalence of this disease is reported only in some specific geographical regions of India. Hence, the aim of this study was to screen the population of Garhwal for beta-thalassemia (β-thalassemia) and thus find out the prevalence in the inhabitants through molecular characterization. For this study, 4,081 individuals were considered, out of which only the ones with elevated HbA_2 levels (64) were subjected to molecular characterization. Mutational studies were carried out for the five most common mutations prevalent in the Indian subcontinent, that is, IVS 1-5 G-C, IVS 1-1 G-T, Codon 41/42 (-TCTT), Codon 8/9, and 619 bp deletion. The present study reports a frequency of 0.5% for β-thalassemia mutations among the subjects we have studied. The analysis of mutation spectrum revealed highest prevalence for IVS-1-5 (G-C) (18.75%) followed by Codon 8/9 (12.5%) and IVS-1-1 (G-T) with 6.25%. Codon 41/42 (-TCTT) and 619 bp deletion were found to be absent in our study population.

Keywords: Beta-thalassemia; Molecular characterization; Mutation, Beta-globin gene, India

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1. Introduction

Thalassemia is inherited, meaning that at least one of the parents must be a carrier of the disorder (Bajwa & Basit, 2022). It is caused by either a genetic mutation or a deletion of certain key gene fragments. Thalassemia patients have fewer than normal hemoglobin molecules. Red blood cells can carry oxygen because of hemoglobin. Anemia and fatigue are major symptoms of thalassemia, so patients of thalassemia in its severe variants may necessitate frequent blood transfusions. However, unlike thalassemia, in anemic condition, the body does not have enough normal and healthy red blood cells.

Thalassemia was originally thought to be the characteristic of tropics and subtropics but due to migration, it is now becoming a substantial global concern (De Sanctis

et al., 2017). Thalassemia is a group of inherited blood disorder transferred from parents to their offspring in an autosomal recessive manner. It is characterized by the abnormal production of hemoglobin. There are two forms of thalassemia: Alpha and beta. The beta-thalassemia (β-thalassemia) is characterized by the mutations in the beta-globin gene, which leads to reduced synthesis or absence of beta-globin chain and is responsible for causing β-thalassemia disorder in individuals from different ethnic groups. The DNA polymorphism in the β-globin gene shows a considerable amount of variability and form a series of patterns (i.e., haplotypes), which occur at varying frequencies among Asian Indians (Kazazian *et al.*, 1984). For the 1st time, the common seven mutations and their haplotypes in the Indians – Frameshift β 8-9 (+G), Nonsense Codon 15 (TGG-TAG), Frameshift β 41-42 (-TCTT), Frameshift β 16 (-C), IVS-1-5 (G-C), 619 bp deletion, and 13 and 25 nucleotide deletion, at 3' end of the gene were reported by Kazazian *et al.* team in the year 1984. According to them, deletion of 619 bp was found to be a common mutation in the Asian Indians. Recent studies reveal that each year between 300,000 and 400,000 babies is born with serious hemoglobin disorder in low- or middle-income countries, out of which 23,000 account alone for β-thalassemia major with about 90% of these births (De Sanctis *et al.*, 2017).

The carrier frequencies of β-thalassemia vary in different parts of the world from 1 to 20% or they may be even higher in some cases (Black *et al.*, 2010). In India alone, the numbers of β-thalassemia patients are approximately 30 million. About 10% of the total world thalassemic individuals are born in India every year (Bashyam *et al.*, 2004). Certain communities in India, such as Sindhis (Colah *et al.*, 2010), Guajarati's (Bhukhanvala *et al.*, 2013), Punjabis (Grow *et al.*, 2014), and Bengalis (De *et al.*, 1997), are commonly affected with β-thalassemia and the incidence varies from 1% to 17% (Gupta *et al.*, 2003). As far as in studies conducted in different parts of the world, more than 150 – 200 mutations causing β-thalassemia have been reported (Cao & Galanello, 2010). While studies conducted in India have identified about 28 mutations in Indian populations (Old *et al.*, 2001), out of these 28 mutations, five to six mutations are found to be common. These include IVS 1-5 (G-C), 619 bp deletion, IVS 1-1 (G-T), Frameshift 8/9, 41/42, and Codon 15 (Bandyopadhyay *et al.*, 2004). The type of mutation varies across different ethnic groups (Bashyam *et al.*, 2004). The frequency of mutations in carriers varies from region to region with the predominant mutation being IVS 1-5 (G-C) (Agarwal *et al.*, 2000), which reflects the ethnic and genetic diversity of populations. The heterogeneous populations belonging to the Indian subcontinent have been widely studied (Grow *et al.*, 2014), by various researchers in Pakistan (Usman *et al.*, 2009), Sindh (Jawahirani *et al.*, 2007), Punjab (Garewal & Das, 2003), Gujarat (Bhukhanvala *et al.*, 2013), Tamil Nadu (Colah *et al.*, 2009), Maharashtra (Ambekar *et al.*, 2001), and Kerala (Edison *et al.*, 2008). However, baseline data on thalassemia were completely unavailable in the populations of Garhwal region. Since the Garhwal Himalayan region is still unexplored due to the unpredictable climatic and weather conditions, the remoteness and location of the place make the research unfeasible in such an area. Individuals are limited to their daily needs and there is a lack of knowledge and education among the public in the Himalayan region. Therefore, considering the severity and the importance of the disorder in affecting people's health and well-being, there is a great need to carry out such studies in the population of this area.

2. Materials and methods

The present research was carried out on 4,081 (2,956 females and 1,125 males) unrelated individuals, from June 2016 until August 2019. The participants constituting the study were 0 – 60 years of age and had no chronic illness or history of infection. The ethical clearance for the present research was obtained by the Institutional Ethical Committee (I.E.C.) of H.N.B. Garhwal University before the commencement of the study.

Demographic information such as age, sex, marital status, occupation, prior history of infection, or chronic illness or surgery were obtained directly through face-to-face interview. Only after a written consent was obtained from the individuals, further study was carried out on the willingly participating ones.

Blood samples (whole blood) for the present study was collected from the district hospitals, public health centers, villages, schools, and local people residing in the five major districts of Garhwal region, including Chamoli, Pauri, Rudraprayag, Tehri, and Uttarkashi. A total of 5 ml of blood were collected from each sampled individual. The blood samples were collected in Vacutainer containing ethylenediaminetetraacetic acid as anticoagulant. Twenty microliters of anticoagulated blood were utilized for hemoglobin analysis. NESTROFT test (Naked Eye Single Tube Red Cell Osmotic Fragility Test) was done using 10 µl of blood, following the protocol described by Thomas *et al.* (1996). Approximately 2 ml blood was utilized for HbA_2 analysis using high-performance liquid chromatography – HPLC (Variant BioRad 2).

Out of total 4,081 blood samples analyzed for hemoglobin and NESTROFT, only 648 individuals were found NESTROFT positive. Further screening for HbA_2

was done for all 648 individuals. Molecular investigation was done only for 64 individuals out of the total 648 screened for HbA_2, as their HbA_2 levels were more than 3.5%. The cutoff level of more than 3.5% of HbA_2 is considered as the gold standard for β-thalassemia trait (Ou *et al.*, 2011). All these 64 individuals were, thus, subjected to the detection of mutation through the amplification-refractory mutation system-polymerase chain reaction (ARMS-PCR), which is a simple method for detecting any mutation involving single base changes or small deletions. ARMS is based on the use of sequence-specific PCR primers that allow an amplification of test DNA only when the target allele is contained within the sample (Little, 2001). Molecular analysis for the blood samples was carried out at the Central Molecular Research Laboratory of SGRRIHMHS, Dehradun, India. Genomic DNA from the blood samples was extracted using QIAamp DNA Blood Mini Kit (Qiagen, Valencia, CA, USA).

The amplification of the targeted sequences of the extracted DNA was carried out through ARMS-PCR technique. The five most common mutations prevalent in India, that is, IVS 1-5 G-C, IVS 1-1 G-T, Codon 41/42 (-TCTT), Codon 8/9, and 619 bp deletion were considered for detecting mutations. The PCR reaction was carried out with initial denaturation at 94°C for 5 min, followed by 30 cycles of denaturation at 94°C for 1 min, 1 min annealing at 60°C, extension at 72°C for further 1 min, and final extension for 5 min at 72°C. After PCR reaction, the products of PCR are obtained in the form of amplicons. To analyze the PCR products, 10 µl of amplicon was loaded on 1.5% agarose gel along with 1 µL of 6X DNA loading dye (ML015). In a separate well 3 µL of 50 bp DNA ladder (MBT084) was loaded as marker. The gel was then allowed to run for about 15 – 20 min at 125 volts and the results were recorded in Gel Documentation System.

3. Results

The distribution of the five mutations, that is, IVS 1-5 G-C, IVS 1-1 G-T, Codon 41/42 (-TCTT), Codon 8/9, and 619 bp deletion, among the 64 individuals was studied using ARMS-PCR. The most frequent mutation was IVS 1-5 (G-C), which was present in 12 study subjects, accounting for 18.75% of the study subjects. While Codon 8/9 was found in 12.5% of individuals, IVS 1-1 (G-T) was found to be present in 6.25% of the study population. On the other hand, 619 bp deletion and Codon 41/42 (-TCTT) mutations were absent in our studied population. The allelic frequency for all the three mutations, IVS 1-5 (G-C), IVS 1-1 (G-T), and Codon 8/9, was found to be same at 0.99. The distribution frequency of the five investigated β-thalassemia mutations is shown in Tables 1-4 according to the four major zones of India in comparison with the previous reports on β-thalassemia in different regions of India.

4. Discussion

Numerous studies on thalassemia have been carried out throughout the globe. India, being a part of the thalassemic belt, faces various problems, such as unawareness, lack of finance, resources, and social stigma for pre-marital screening (Mohanty *et al.*, 2013; Verma *et al.*, 1997). Despite the enormous amount of research and rapid developments seen during the past decade, it still continues to be a national concern. According to the World Health Organization (WHO), the total thalassemic reported in India alone accounts for 80 – 90% and is a common hemoglobinopathy (Haritha *et al.*, 2012). β-thalassemia is considered to be the cause of morbidity and mortality along with the source of economic burden to the community (Piplani *et al.*, 2013). As per the WHO (WHO, 2001) records, β-thalassemia is a common hemoglobinopathy in India, and therefore, numerous studies have been carried out for Indian population for the distribution of thalassemia mutation. Since India represents an extremely heterogeneous population with numerous tribal pockets, diverse racial origin, and high inbred diseases frequency among certain communities, the prevalence rate of thalassemia is very high for some particular communities. There are diverse ethnicities residing in different parts of the Himalayan belt. A study carried out in western part of India, conveyed 22.7% thalassemia carrier women diagnosed with anemia (Mulchandani *et al.*, 2008).

Molecular confirmation was done to ascertain authentic reporting. In different local communities of India, the reported range for the five most common β-thalassemia mutations was between 0.3% and 17% (Agarwal & Mehta, 1982; Weatherall & Clegg, 2001a; 2001b; WHO, 2008). The present research reported a frequency of 0.5% for β-thalassemia mutations, which is within the range described for the Indian population. The analysis of mutation spectrum revealed the highest prevalence for IVS1-5 (G-C) (18.75%) followed by Codon 8/9 (12.5%) and IVS 1-1 (G-T) (6.25%). The frequency of mutation was compared with the other reported mutations of Indian population (Tables 1-4). Colah *et al.* (2009) discovered IVS 1-5 (G-C) as a pre-dominant mutation throughout India, with 44.8% prevalence in the north and 71.4% in the east. This pre-dominancy is in unison with our study where the frequency of mutation was found to be 18.75% for IVS 1-5 (G-C).

In contrast to our study, a higher frequency (88.6%) of IVS I-5 (G-C) was reported in Orissa, 78.8% in Andhra Pradesh, and 77.2% in West Bengal (Colah

Table 1. Distribution of five most common mutations for beta-thalassemia among the eastern zone of Indian population

State	Sample size	619 bp deletion	Co 8/9 (+G)	Co 41/42 (-CTT)	IVS-1nt 5 (G-C)	IVS-1nt 1 (G-T)	References
West Bengal	2,456	1.0	0	4.0	77.2	4.0	Colah *et al.* (2009)
	11	45.450	0	0	0	0	Bashyam *et al.* (2004)
East India	80	0	0	17.50	28.75	0	Kukreti *et al.* (2003)
Bihar	2,456	0	0	15.0	70.0	0	Colah *et al.* (2009)
Jharkhand	2,456	0	0	0	75.0	0	Colah *et al.* (2009)
Orissa	2,456	1.2	0	2.6	88.6	0	Colah *et al.* (2009)
	276	0	0.38	0	4.59	0.38	Sahoo *et al.* (2014)
Bihar, Chhattisgarh, and Jharkhand	1,642	0	3.6	5.4	51.8	0	Nagar *et al.* (2014)

Table 2. Distribution of five most common mutations for beta-thalassemia among the western zone of Indian population

State	Sample size	619 bp deletion	Co 8/9 (+G)	Co 41/42 (-CTT)	IVS-1nt 5 (G-C)	IVS-1nt 1 (G-T)	References
Maharashtra	2,456	1.7	1.7	2.0	66.5	0.7	Colah *et al.* (2009)
	126	2.38	6.34	3.96	65.07	9.52	Satpute *et al.* (2012)
Gujarat	2,456	6.5	12.6	6.9	48.6	5.0	Colah *et al.* (2009)
Gujarat (Sindhi)	1,233	49.2	0	0	0	25.5	Vaz *et al.* (2000)
Gujarat (Lohanas)	1,233	0	0	0	0	31.2	Vaz *et al.* (2000)
Gujarat (Punjabis)	1,233	0	0	0	0	34.7	Vaz *et al.* (2000)
Goa	2,456	0	0	0	15.4	0	Colah *et al.* (2009)
Rajasthan	2,456	1.1	2.3	9.9	72.4	3.3.	Colah *et al.* (2009)
Madhya Pradesh	2,456	10.6	19.9	5.7	36.9	11.3	Colah *et al.* (2009)
Chhattisgarh	2,456	23.5	23.5	0	35.3	5.9	Colah *et al.* (2009)
East-west population							
Rajasthan, Maharashtra, Assam, and West Bengal	75	13.23	0	0	0	0	Shah *et al.* (2017)

Table 3. Distribution of five most common mutations for beta-thalassemia among the southern zone of Indian population

State	Sample size	619 bp deletion	Co 8/9 (+G)	Co 41/42 (-CTT)	IVS-1nt 5 (G-C)	IVS-1nt 1 (G-T)	References
Andhra Pradesh	2,456	0	0	0	78.8	0	Colah *et al.* (2009)
	39	0	0	0	76.92	0	Bashyam *et al.* (2004)
Karnataka	2,456	0	0.8	0.8	67.2	1.6	Colah *et al.* (2009)
	28	0	0	0	64.28	3.57	Bashyam *et al.* (2004)
Kerala and Tamil Nadu	2,456	0	0	6.31	56.3	0	Colah *et al.* (2009)

et al., 2009). The previous studies showed that Codon 8/9 had a higher prevalence of 37.2% in immigrants from Pakistan followed by 23.5% in Chhattisgarh population (Colah *et al.*, 2009), whereas in our study, it was found only 12.5%. Codon 41/42 (-TCTT) was found to be higher (17.50%) in East Indian population in the previous studies (Kukreti *et al.*, 2002) and was also reported in Punjab and Haryana (13.5%) (Colah *et al.*, 2009), but for our study, the frequency of this mutation was nil. A study carried out in Himachal Pradesh/Jammu Kashmir region by Colah *et al.* (2009), reported 37.5% IVS 1-1 (G-T) mutation. The very same mutation is found to be the second highest among the Punjabi Hindu population of Gujarat (34.7%) followed by Lohanas of Gujarat (31.2%) (Vaz *et al.*, 2022). The previous

Table 4. Distribution of five most common mutations for beta-thalassemia among the northern zone of Indian population as well as Garhwal population

State	Sample size	619 bp deletion	Co 8/9 (+G)	Co 41/42 (-CTT)	IVS-1nt 5 (G-C)	IVS-1nt 1 (G-T)	References
Punjab, Haryana, and Uttar Pradesh	1,050	21	12.1	8.7	34.1	15.8	Verma *et al.* (1997)
Delhi	46	34.8	13	9.8	22.8	19.6	Madan *et al.* (1998)
Uttar Pradesh	376	2.5	0	0	64.3	0	Agarwal S *et al.*, 2000
	98	12.5	5.20	6.25	54.0	3.12	Agarwal S *et al.*, 2003
	48	0	21	4	46	12	Christopher AF *et al.*, 2013
Uttar Pradesh	2,456	0.8	1.6	9.6	64.8	5.6	Colah *et al.* (2009)
Jammu, Kashmir, and Himachal Pradesh	2,456	0	0	0	50.0	37.5	Colah *et al.* (2009)
Punjab	780	18.7	14.8	15.3	31.8	12.8	Garewal & Das., 2003
	997	22.47	16.35	10.83	24.97	14.44	Black *et al.* (2010)
Punjab and Haryana	2,089	9.6	7.7	13.5	28.8	17.3	Panigrahi & Marwaha (2007)
Chandigarh (Sindhi)	87/1233	0	0	0	25.5	0	Panigrahi & Marwaha (2007)
Chandigarh (Punjabi)	87/1233	0	0	0	34.7	0	Panigrahi & Marwaha (2007)
Chandigarh (Lohanas)	87/1233	0	0	0	31.2	0	Panigrahi & Marwaha (2007)
Other North Indian States	62	94.1	0	0	0	0	Chakrabarti *et al.* (2005)
Uttarakhand							
Garhwal (Uttarakhand)	4,081	0	12.5	0	18.75	6.25	Present study

studies (Baig *et al.*, 2006) reported a high frequency of 37.3% for IVS 1-1 (G-T) mutation in Punjabi population residing in Pakistan. On the contrary, our study exhibited only 6.25% of the screened population having IVS 1-1 (G-T) mutation. In our study, Co 41/42 (-CTT) and 619 bp deletion were completely absent.

Of the total 64 individuals who were screened for mutational spectrum, mutations were found only in 24 individuals and the remaining 40 individuals did not reveal the presence of any of these five common mutations. The remaining 40 individuals may possess a novel mutation or can only be carrier. The confirmation of novel mutation can be done through DNA sequencing. However, DNA sequencing for the study presented could not be carried out due to economic constraint and limitation of resources. Furthermore, only 64 individuals out of the total 648 screened for HbA_2. The small sample size may suffer from some biases in our findings and thus the cautions are needed in interpreting our findings.

Despite some limitations, our study could have important implications. There are various programs implemented by the state governments and non-governmental organizations (NGOs) around the world, including India, for the awareness of thalassemia and also about its fatal effects. However, nothing has been done to make the population of Garhwal aware of this disorder. In the developing country like India, thalassemia is not rare, and our nation is not an exception. In Pakistan, a bill is passed by the government where it is mandatory to carry out carrier testing for relatives of thalassemia patients. A similar system of bill is also placed in Dubai, Saudi Arabia, and Abu Dhabi where the frequency of beta-thalassemia is 19% (Cao & Kan, 2013). All these together with our findings signal the importance of initiatives and implementations of some targeted intervention programs in our study areas.

Furthermore, some caveats are noteworthy. Consanguineous marriage causes clustering of the mutations among some of the communities and, therefore, should be avoided. Clinical management, genetic counseling, and prenatal diagnostic techniques should be made available to the individual level. Awareness programs regarding anemia and thalassemia must be implemented for the general public through media and other modes, to make the public aware about this deadly disorder. As, the burden of thalassemia can be reduced at community and country level only through awareness, screening, and prevention strategies in conjunction with each other (Kumar *et al.*, 2015).

5. Conclusions

It is the first study to report and provide baseline data on the prevalence of β-thalassemia among the studied population of Garhwal, India. This research exhibited prevalence rate

of 1.5% for beta-thalassemia trait (BTT) on the basis of elevated level of HbA_2 of ≥3.5%. The present study reported a frequency of 0.5% for β-thalassemia mutations. The analysis of mutation spectrum revealed highest prevalence for IVS 1-5 (G-C) (18.75%) followed by Codon 8/9 (12.5%) and IVS 1-1 (G-T) with 6.25%, whereas Codon 41/42 (-TCTT) and 619 bp deletion were found to be absent.

The present research is a preliminary attempt to record the prevalence rates of thalassemia, prior for Himalayan region of Uttarakhand. The study suggests that emphasis should be laid down on the implementation of population screening programs to reveal the exact number of individuals carrying the gene for BTT and having anemia. The information provided in this research can be used for planning population-based mutation screening strategy along with pre-marital screening.

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Conflict of interest

The authors have no conflicts of interest to declare.

Author contributions

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Ethics approval and consent to participate

The ethical clearance for the present research was obtained by the Institutional Ethical Committee (I.E.C.) of H.N.B. Garhwal University (2016/01). Written consents were obtained from all the participants.

Consent for publication

Not applicable.

Availability of data

The datasets used and/or analyzed during the present study are available from the corresponding author on reasonable request.

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RESEARCH ARTICLE

Identifying resources for promoting healthy aging in community

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Abstract

Community-dwelling older adults depend on resources for stability and safety as they age. This study explores resources that mature over time to improve the quality of aging in community (AIC) within communities that are not modeled for aging populations. Recommendations for future research and community programs to improve the ability of community members to age in community were made based on the findings. This qualitative study used semi-structured interviews with open-ended questions on 16 older adults who attend programs designed to increase community interaction. According to data gathered, the three most important resources were social, health, and financial. AIC requires multiple resources to sustain basic needs and provide quality living. Early allocation of resources improves the likelihood of successfully AIC.

Keywords: Aging in community; Gerontology; Retirement planning; Village model

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1. Introduction

Dependency on resources for stability and safety increases as humans age, especially for those who wish to age in typical community environments. Aging community models allow older adults to remain in one community by optimizing resources allocation and distribution. Alternatively, typical community models are designed for members who gather and use their own resources. However, in this model, access to available resources may be insufficient to meet basic individual needs or keep up with community standards. Therefore, one may result in a deficient quality of living for that community. Utilizing village model organizations can offer support for aging in community (AIC). However, knowing what is needed in preparation for this lifestyle can prevent insufficiencies before entering the later stages of aging. This study explores resources that mature over time to improve the quality of AIC within communities that are not modeled for aging populations. Recommendations for future research and community programs to improve the ability of community members to AIC can be suggested in this study finding.

1.1. Literature review

AIC is the preferred environment for older adults in the United States (Cutchin, 2003). Adults continue to live in their own communities and homes, and there is no requirement

for age, income, or ability level like purposefully designed aging communities (Tovel & Carmel, 2014). To AIC, both personal resources for reacting to unforeseeable events and preparation for future events can influence the likelihood of stability and success for someone to AIC (Hossain & Ismail, 2020). Resources are a supply that someone can use in a time of need and be able to assist them in working through a situation (Merriam-Webster, 2021; Szanton *et al.*, 2014). More specifically relating to the study of AIC, resources refer to support measures required for adults during daily activity that would otherwise be supplemented by staff at adult day cares or assisted living facilities (Cutchin, 2003). A *theta* refers to the decomposition of a resource compared to the resources forecast over time (U.S. Census Bureau, 2006). The theta, or the forecasted decomposition, of resources for aging represents the allocation of and ability to capitalize on resources as someone approaches needing to depend on this resource for stability (U.S. Census Bureau, 2006). Resources preparation in the context of aging is battling a negative *theta*. Therefore, as we age the ability to progress positively in resource allocation becomes less likely.

As resources and abilities decline, the safety of the aging person also declines, and the resources for recovery are diminished (U.S. Census Bureau, 2021). While AIC, homeowners are less likely to want to leave their residence as they age. Only 5.3% of citizens over the age of 65 relocated in 2006 (Di, 2003; Golant, 2008). Therefore, as people age, they are likely to remain in housing structures that are older and less likely to remain current on proper maintenance (Lagergren *et al.*, 2017). The allocation of financial resources at this stage becomes less liquid as a significant source of net worth for older adults sits in the equity of their home (Park *et al.*, 2016). As adults age with financial resources locked in assets, releasing these assets may cause additional burden on their daily life.

A reduction on one resource poses potential access limitations to other resources. This includes access to goods and services which can be a determinant of longevity of life (Hughes *et al.*, 2008; Steverink *et al.*, 2001). Resources used while AIC impact the quality of life one wishes to have. Financial resources can control the level of access to goods and services which directly impact one's ability to set their level of comfort or meet basic individual needs. Utilization of resources can also have compounding effects. Social networks take time to develop and have a direct impact on other accessing or building resources for aging adults (Taylor & Doverspike, 2003). However, resources also differ in the ability to use with liquidity. For example, one cannot access their home equity without risking financial stability, but they can call on a community member for assistance and still retain social stability (Bookman, 2008). As adults age, it is important to have adequate available resources. As we age, we must begin creating these resource caches. The bigger the contribution is, the more likely one will have adequate resources when retirement status begins and aging impacts resource utilization (Hou, 2019).

Recent studies in gerontology have identified programs that are directing attention to providing supplemental support for older adults within their existing communities (Greenfield *et al.*, 2012; Hou, & Cao, 2021; Hou *et al.*, 2019). The village model is a community collection of services that are governed by a legal entity, which can assist in aiding people in AIC. The village model can offer civic engagement and empowerment for the members to reduce AIC burdens while improving quality of life (Sharpe, 2021). Village models also address resource dependency through its design which allows members to access needed services with reduced or no dependency on their individual resource caches (Rameli & Marimuthu, 2018).

1.2. Significance of the study

While longitudinal studies emphasize the need for preparation (Gibbs, 2007), determining which resources are most desirable and its impact on older adults' quality of life is difficult to predict. Many preparation programs focus on finite aspects, such as retirement income (Yu *et al.*, 2021), but fail to recognize impacts on quality of life. To determine what preparation methods are likely to lead to a more preferred outcome of AIC, we must understand what areas are most important to older adults that require long-term design. We can then recommend further research that emphasizes a quality lifestyle while living in community. This study intends to determine what quality-of-life resources require long-term investing for individuals that desire to AIC.

2. Data and methods

This qualitative study gathered information from older adults who attend programs designed for them to interact in their communities. Participants were selected because of their membership of one of two social groups, the lifelong learning program (LLP) and a village program. Both community models offer similar services to aid their members to promote AIC. The village model is a collaboration of community members, volunteers, sponsors, and a governing board that hosts an array of services and activities for its members. The services can include access to volunteers for assistance maintaining their home and lifestyle such as handyman services, transportation service, financial guidance, and more. The village model is often associated with a non-profit organization to support the needs of the members (Greenfield *et al.*, 2012). The LLP functions very similarly to the village model but is

associated with a learning institution or university. The LLP may bring additional activities that support continuing education throughout the lifespan to promote psychological and emotional function. All participants are currently AIC and have entered retirement status. A total of 16 members participated in the study; seven were from the LLP and nine were from the village program. The distribution of the sample populations demographics is listed in Table 1.

Data were collected by a semi-structured interview using open-ended questions. Responses were separately analyzed for thematic consistencies to determine what resources affect respondents' daily interactions and activities. An effect on daily interactions or activities will be noted if the response indicates that the resource hinders or assists in the ability to participate. Interviews consisted of 21 questions designed to stimulate responses about the daily lifestyle of participants that participate in AIC. Questions target the participants daily life activities, daily needs, daily interactions, long-term planning, and interactions that occur. Questions were open ended to promote spontaneous responses. Interviews were audio recorded, transcribed, and read repeatedly by different raters for analysis. Meaningful words, phrases, and sentence topics were extracted and compared to other participants' codes for each question (Slingerland *et al.*, 2007). Data were analyzed by each study group and as a collective whole. Initial codes were generated, and common thematic occurrences were analyzed for responses. Codes generated focused on the main emphasis which the responses were targeting. The codes were then filtered to represent the resources which impacted the statement. These resources were represented by their use, desire to use, reliance on, notice of deficiency, or total absence of. Codes were discussed between investigators to ensure that the central theme of the responses was represented by the codes used. The codes applied to each response were reviewed and agreed on by investigators.

3. Results

Several themes occurred throughout the open-ended questions. Themes were discussed for the impact they have on the participants' perception of quality while AIC and the potential for impact preemptive planning would have on their current and future resources. The themes that provided the most insight for determining long-term resource planning are listed in Table 2. Responses collected provide information that was affected by or correlated to other themes. Therefore, information gathered frequently crossed between multiple coding variables.

Social network represents the participants' interactions with other community members or friends. This code does not reflect interactions with family, only interactions with community members or close friends. Health represents the participants' interactions with attending to their personal health-care needs, for example, attending a health care-related appointment, following a health-care regimen, or activities that affect or are affected by their health status. Financial condition represents the influence on finances for interactions or decision-making. The code represents limitations, supportive advice, personal privileges, and reflections of previous actions related to finances. The above table delineated the total account of code usage for each sample group and the total number of uses for both groups. The table also represents the percentage of answers which the codes occurred in. The distinction of the columns represents the frequency of occurrence for the code's use within the variety of questions asked but also represents the intensity in which the responses were reflecting on the use of that code.

The most commonly used code in frequency and intensity was social network. While the health code appeared as a response for the same number of questions, the participants provided more depth in the responses when the social network code was used. This persists in both groups. The financial code referenced one additional count over the health code; however, the range of responses which it occurred was less. Other codes were used in the analysis of the data but did not indicate a significant occurrence of frequency or intensity. Therefore, those codes were not able to provide sufficient data for analysis.

3.1. Social network

Socialization was noted as an important aspect of daily life and responses indicated that it increased in importance as we age. The programs the participants were involved with assisted in filling this need for socialization. "It encourages me to get out of my house and interact with people," responded one participant when asked about the impact of the program. Socialization through the village program was also stated to have benefited the community for connecting people for personal socializations. The socialization theme noted impacts on quality of life within the community as well. One participant stated, "I go out and I meet people at the supermarket that I know from (village program), you know."

3.1.1. Lacking socialization

Adversely, the participants also noted that there is a generational change in which the younger generations are not socializing in typical manners. This causes the participants to presume that there is a disconnect from younger generations. Responses noted that younger generations use technology as a means of socialization,

which yielded mixed feelings among responses. Some participants noted the amazement of what younger generations can do with technology and socialization. Others remarked on how technology and the abundance of its use distanced the younger generation from each other and limited their ability to be social.

3.1.2. Accessing socialization

Transportation abilities were expressed in several areas of responses from participants which reflected limitations in social networking. Commonly, barriers for aging adults were indicated by transportation being a limiting factor to get to events and meet up with others. It was openly expressed that services are needed to maintain active living among those AIC so that they can remain active in daily tasks. A common notion was the need to keep moving to avoid the status of being "old." When asked if the participants consider themselves old, a common response was that being old was a state of mind and that they keep pushing the concept of being old further away as they age. The defining moment of being old for those participants was when a person stops doing things. "…if you let yourself get old then you are going to be old. You're just going to stop. Once you sit down then that's it. You've got to keep moving and keep going and try to stay sharp…" expressed the participant. The theme of being active and part of activities requires transportation to prevent this limitation from occurring. However, in general, the interactions remarked on the need to retain socialization as a form or retaining quality of life.

3.2. Health

When discussing matters that impact the daily lives of the respondents, the most frequent remark was that of health, health care, mental health, and maintaining a healthy lifestyle. The importance of health maintenance and having an early start on healthy lifestyles was emphasized as a driving force of daily life and daily planning. One participant stated, "You have to do stuff actively to give yourself the best chance to remain physically healthy and intellectually stimulated." Another respondent stated, "Be open to new experiences, take care of your health, and get exercise." While other responses did not directly state that health was the factor, they did indicate by exemplar that health plays an important role in aging such as the statement "enjoy life and especially for my grandkids who are young, enjoy being a kid now cause you're a kid for like maybe 12 – 13 years that you're actually a kid. That you can run and do somersaults and act goofy. Enjoy it now cause it's gonna be gone and you're gonna wish you could go back cause sometimes I wish I could relive when I was younger."

3.2.1. Health influence

Health-related responses influenced other categories as well. For this reason, personal health was the most referenced thematic code from all respondents. The ability to participate in events was impacted by health-related events such as attending frequent doctors' appointments or planning their schedule to suit upcoming procedures. When asked for advice to future generations, remaining physically and mentally healthy were expressed in almost every response. The responses suggest that your physical and mental health is a high priority for successful aging.

3.2.2. Health activity

Exercise was indicative of retaining health status and ability to participate in events. Participant responses signaled that a status of aging and being labeled as "old" was highly reflective of physical abilities. Responses indicated that when you are no longer active, you have progressed to status that they would consider "old." Additional limitation of physical ability was in reference to ambulation or ability to climb stairs. Respondents also indicated that weight loss plays a part in addressing physical limitation, noting that previous weight loss had a positive effect on their ability to be self-ambulatory, or the need to lose weight could increase physical abilities.

Additional dialog reflected the need for increased maintenance services for aging adults. While this is both physical and financial, respondents did not remark on the price of maintenance being an impact, but that it was needed to remain at their current level of living. The respondents indicated the need of maintenance as a service for aging within their home/community in the form of retaining a healthy environment. This suggests that the need for maintenance has physical limitations for an aging individual and impacts their environmental health.

3.3. Financial conditions

Responses in which financial resources were noted had the most detail among the thematic codes collected. Finances were discussed frequently among responses as areas of limitation, need of support, desire for improvement, desire for education, and suggestions for future generations. Participants remarked that they were subject to daily limitations because of funding. Multiple participants mentioned that finances directly impacted the integrity of the programs because it could exclude less financially stable members. They stated that membership fees, cost of activities during events, and cost of transportation could be a limiting factor for members to become part of their social network, thus limiting their engagement with other members of the community. In addition, participants from

Table 1. Sample population demographics

	Village program	Lifelong learning program	Total	% of samples
Gender				
Male	4	3	7	43%
Female	5	4	9	56%
Marital status				
Married	5	5	10	62.5%
Widowed	3	1	4	25%
Single	1	1	2	12.5%
Age (years)				
60 – 69	1	3	4	25%
70 – 79	4	3	7	43%
80 – 89	3		3	18%
90+	1		1	5%
Family				
Children			12	75%
Grand children			7	43%
No children			4	25%

Table 2. Thematic codes distribution

Thematic code	Village program	Lifelong learning program	Total count	Percent of questions codes found in
Social network	32	15	47	36.8%
Health	6	10	16	36.8%
Financial condition	11	6	17	31.5%

both groups indicated that early financial planning was important, specifically knowledge of proper budgeting to prepare themselves while on a fixed income. One participant suggested that future generations should "make sure that as hard as it is, put money away and save it."

3.3.1. Careers

Career planning, advancement, and quality were also discussed throughout the interviews. Participants noted that early career planning, or lack of, was impactful on how their financial resources were affected in the present day. One participant remarked "there's just not enough information provided to kids to direct them." Another noted that exposure should be increased, stating "I think in high school they could have more speakers coming in talking about what they do for a living and their jobs." Participants expressed that it is as important to be part of a career that you enjoy as well as one that provides financial stability. In addition, they remark that the ability to change careers or advance to more important roles should be encouraged to the future generations.

3.3.2. Education

While some participants were promoting the idea of continuing education, caution on how education was obtained differed greatly. Some participants noted that loans for education were not suggested because of their negative long-term financial impacts, but college in general is very beneficial. Other participants noted that technical learning, either by school or apprenticeships, was preferred methods of developing career skills because of their positive financial impacts. As a common point, the participants agreed that the education path that the future generations consider should also include an understanding on how much the cost of the education will be compared to the potential income from that career. For example, one participant noted that some college degrees cost more than 2 years' salary for the job they are seeking, and therefore, the student's ability to start becoming financially stable is delayed.

Finally, on multiple occasions, participants noted that they have experienced or are currently in a state where financial advisory is needed. The general lack of financial experience and understanding was indicated among multiple participants in response to multiple questions. Participants indicated that increased financial awareness and early planning would have a heavy impact on the function of their status.

3.4. Differences among the two sample groups

3.4.1. Village program

Responses of the village program group showed a tendency to focus on socialization and community. The responses from participants often reflected how socialization was a top priority of their daily lives. The responses that included socialization occurred before offering other information that the question prompted for. The sense of community in the village program group also resonated throughout the body of their responses. The group tended to remark on how they were able to help or be helped by others within their community and groups. The use of their program appeared to promote the acquisition and continuance of relationships with other members. This, in turn, also supported the health of the model by promoting active living and assistance to attend health appointments such as transportation or chaperons.

When referring to less preferred aspects of the group, the village program had several respondents note cultural status as a negative affiliation with the group. One member remarked "So, it's hard for somebody who is not

bilingual, and English isn't their native tongue…." Another respondent felt that it was important to remark on the race of a caregiver "She has somebody that comes in 6 h a day, a blackwoman, but I really got to like her intensely." These cultural references suggest that the village program population may not be a heterogeneous representation of that community but more of a defined set of demographics among that community.

3.4.2. Lifelong learning program

Responses for LLP focused on continued self-improvement and independence. LLP members often remarked on personal well-being by referencing the model's ability to retain mental and emotional health. Responses reflected that the LLP members were attempting to improve their knowledge or expand their understanding of life skills, such as finances or personal health. While socialization was mentioned, it was not as highly promoted for this group.

A notable aspect of LLP is the similarities in the respondents' dislikes. They often referred to the activities that they were part of, particularly the presentations, and how the members take the activities very seriously. This group participated in activities with high regard and remarked on when activities had lackluster performance, or a belittling topic was offensive to the members. One response referenced a presentation, "They're speaking down a little bit…Why would I want to say something bad about someone? [So I] just let it go." Consistently, the respondents remarked that they have preferences of topics but were open to variety. Responses often indicated that they were pleasantly surprised by presentations with new categories that turn out to be interesting to them. However, the speaker's presentation of the material was paramount to their perception of the activity.

4. Discussion

Evaluation of resources through qualitative methods reveals several areas with potential long-term attributions. The complexity of their interactions should not be underestimated but the focus of this investigation is to open the dialog of what resources are perceived as important and what the hierarchy of importance was. Several assumptions were made about resource allocation and distribution that would impact the ability to AIC. The implications of financial resources, as gathered from the data, was that long-term preparation for income, investments, and utilization were heavily impacting the ability for the participants to AIC. Personal health and well-being resources were noted to increase opportunity for successful AIC by minimizing complications that can lead to the need for intervention. Finally, social resources impacted the ability to compromise when faced with complications that occur when AIC.

4.1. Social resources

Social resources have the most direct impact on maintenance and security of AIC. While financial and health resources can directly impact access, social resources had the most impact on the quality of life while aging. Responses describe the need for participants to retain access to socializing for their mental and physical health. The ability to interact with others, whether through planned events or by serendipity, was among the most expressive responses. The notion that a person will become old because they are no longer participating in activities was expressed by multiple participants and thus emphasizes its importance. This can directly impact mental health by addressing isolation. Isolation is also linked to an increase in cognitive decline among those in retirement (Barbosa *et al.,* 2016). Isolation aside socialization among retirees promotes physical health through activities. While physical activity is expected to decline over time, social opportunities can combat this decline (Gillsjö *et al.*, 2021).

4.2. Health resources

Health resources also provide variability to the individual because of predispositions of medical conditions. However, general health allocation is indicated to still impact successful AIC because of peripheral factors. Health issues have limited participants' abilities to complete tasks that assist in retaining quality of life. Such tasks include general ambulation, home maintenance, cooking, and cleaning. Health resources limiting the level of activity, mobility, or strength can force participants to increase the *theta* of their health resources to accomplish more basic tasks, such as hygiene and health monitoring. If early health maintenance occurs, there is an increase in health reserve and a reduction in depreciation occurs. In addition, physical health is found to increase a person's perspective of successful retirement (Hauff *et al.*, 2020). However, the absence of an overall illness or disease does not create a sense of health; an individual must also feel as if they are able to engage in life events and retain a healthy lifestyle (Beier *et al.*, 2018).

4.3. Financial resources

Financial planning for retirement occurs in three stages: Planning, saving, and investment. However, 70% of Swedish adults 18 – 65 have not begun planning on determining what they would need to accomplish to financially retire and only 44% have set aside funds for

retirement (Ihle *et al.*, 2018). This indicates that the most tangible resource for AIC is not commonly considered until after the general age of retirement is surpassed. While resources for AIC vary for each person, the initial stages are forgone, and success becomes directly conflicting with the resources *theta*. To obtain adequate financial resources, planning and execution would require early adaptation and discipline. This was seen in the participants' desire to obtain career choices that were financially sound and did not community the individual underwater with investment. Furthermore, the desire for education on financial planning suggested that participants were also willing to begin the planning stage to protect their futures. While financial investing is indicative of holding finances in positions of growth, participants entering the stages of retirement can still participate in financial planning even if investments may not be as beneficial. This can indicate that financial planning is still an asset after retirement, but the impact of early planning can increase the likelihood of successful AIC. Importantly, those with higher socioeconomic status are proven to believe they have higher probabilities survival in retirement (Choi *et al.*, 2012).

4.4. Social resource development

Socialization resources develop differently than financial and health resources. Socialization resources require proactivity in preparation, allocation, and distribution. This resource is also more difficult to quantify. A person's finances can be measured numerically and propose a rate of depreciation over time. A person's health can be diagnosed and projected based on symptoms and medical history. However, social resources can be difficult to ascertain based on personality and can be affected by events with no direct correlation to the individual. While the participants were part of the same two programs, this does not represent their relationship with other members. Within the programs, members may have different social connections and therefore different access to the benefits of those social connections. In addition, the *theta* of social resources holds more volatility. Miscommunication with a peer could break access to social resources and, depending on other social connections, potentially expand that loss to other social resources. However, if a social connection is longer term, then the likelihood of losing that resource also decreases and the likelihood of interactions increases (Hou, 2020).

Social resources encompass both the acceptance of others into life and the outreach to be part of others' lives. Transportation is an important attribute to be part of social interactions, including medical appointments, social meetings, family events, and religious gatherings. As aging progresses, the likelihood of hazard from self-transportation increases and the dependence on other methods increases. With assistance from friends and family, people are less likely to attempt to self-transport, reducing the opportunity for hazard while also increasing their utilization of social resources (Yuen *et al.*, 2007).

As we discuss the three main categories of resources indicated from participants, it becomes apparent that they are intertwined more than they are independent. Access to health and social resources can be greatly affected by financial resources and the ability to use them with liquidity. In using resources to confront unexpected events, medical episodes can become costly and decrease access to social events, especially if physical impairment persists. Furthermore, socialization that is overindulged or not properly planned for can diminish financial resources or lead to health implications.

4.5. Limitations

This study is limited to the collection of sample groups by method of convenience sampling from the two programs. The sample size is limited by this design, and therefore, generalization to other demographics and geographical areas may not be appropriate. The sample size also limited the ability to determine intergroup demographic implications of the results. While it is known that there is an effect on independence scores and education level, the sample size was not adequate for detecting these variables (Hou, 2020; Yuen *et al.*, 2007). Interview questions were analyzed without retrospective review of the participants and are representative of the research investigators interpretations of responses. Therefore, it is recommended that the future studies should include purposeful focus groups to provide deeper insight into the phenomenon and qualitative data be collected to confirm the results. One strength of this study is that it is the first to attempt to determine the areas of opinionated importance when discussing long-term preparation for AIC. These results were interesting and prompted further inquiry to assist in guiding preparation for successful AIC. Further studies should also direct questioning to determine if participants are aging in the right place (AIRP) with respect to the appropriation of resources. As research pertaining to AIRP suggests that housing should be able to support individual insufficiencies for resources, the determination of resource hierarchy requires continued investigations (Canham *et al.*, 2022).

5. Conclusions

AIC requires an abundance of resources to sustain basic needs and provide a quality of living. Early allocation of resources improves the likelihood of successfully AIC.

According to data gathered, the three most important resources are social, health, and financial. Each resource impacts on a person's ability to remain in community and continue to age. As aging progresses, the use of these three resources begins to increase, causing a stress on the cache. Therefore, higher accumulation of these resources can be reflective of successful AIC.

While other options for aging exist, such as moving in with family or friends, retirement communities, assisted living facilities, and skilled nursing homes, AIC remains the preferred method. While research on current services is valuable to the understanding of this issue, the data collected indicate that longer term plans would be beneficial. This is because of the social health and financial resources required to access these services. A better understanding of these complex and interconnected resources can provide valuable insight into successful AIC.

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Conflict of interest

The authors have no conflicts of interest to declare.

Author contributions

Conceptualization: Adam Reres and Su-I Hou
Formal analysis: Adam Reres and Su-I Hou
Writing – original draft: Adam Reres and Su-I Hou
Writing – review & editing: Adam Reres and Su-I Hou

Ethics approval and consent to participate

This study has been approved as an exempt study by the UCF Institutional Review Board (SBE-17-12893). A cover page with consent information was provided with the paper survey version and "click-through consent page" for the online survey version, before participants voluntarily agreed to take part of the anonymous survey.

Consent for publication

Not applicable.

Availability of data

The data are not publicly available. Please consult the corresponding author for the data access.

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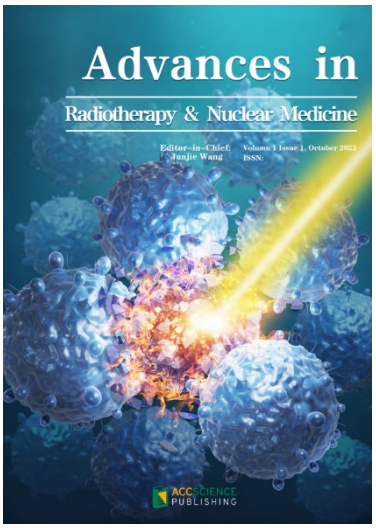

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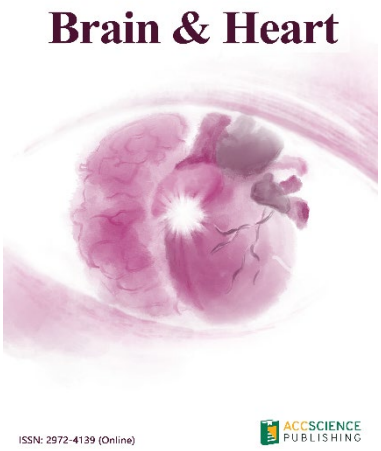

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