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Review Paper

Acceptability of childhood screening: a systematic narrative review

J. Carlton [a, *], H.J. Griffiths [b], A.M. Horwood [c], P.P. Mazzone [a, e], R. Walker [a, f], H.J. Simonsz [d]

[a] School of Health and Related Research (ScHARR), Regent Court, 30 Regent Street, Sheffield, S1 4DA, UK
[b] Academic Unit of Ophthalmology and Orthoptics, The Medical School, University of Sheffield, Health Sciences School, Beech Hill Road, Sheffield, S10 2RX, UK
[c] Infant Vision Laboratory, School of Psychology and Clinical Language Sciences, University of Reading, Infant Vision Laboratory, Earley Gate, Reading, RG6 6AL, UK
[d] Department of Ophthalmology, Erasmus Medical Center, P.O. Box 2040, NL-3000CA, Rotterdam, the Netherlands

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ABSTRACT

Objectives: A systematic narrative literature review was undertaken to assess the acceptability of childhood screening interventions to identify factors to consider when planning or modifying childhood screening programs to maximize participation and uptake.
Study design: This is a systematic narrative literature review.
Methods: Electronic databases were searched (MEDLINE, EMBASE, PsycINFO via Ovid, CINAHL, and Cochrane Library) to identify primary research studies that assessed screening acceptability. Studies were categorized using an existing theoretical framework of acceptability consisting of seven constructs: affective attitude, burden, ethicality, intervention coherence, opportunity costs, perceived effectiveness, and self-efficacy. A protocol was developed and registered with PROSPERO (registration no. CRD42018099763)
Results: The search identified 4529 studies, and 46 studies met the inclusion criteria. Most studies involved neonatal screening. Programs identified included newborn blood spot screening (n = 22), neonatal hearing screening (n = 13), Duchenne muscular dystrophy screening (n = 4), cystic fibrosis screening (n = 3), screening for congenital heart defects (n = 2), and others (n = 2). Most studies assessed more than one construct of acceptability. The most common constructs identified were affective attitude (how a parent feels about the program) and intervention coherence (parental understanding of the program, and/or the potential consequences of a confirmed diagnosis).
Conclusions: The main acceptability component identified related to parental knowledge and understanding of the screening process, the testing procedure(s), and consent. The emotional impact of childhood screening mostly explored maternal anxiety. Further studies are needed to examine the acceptability of childhood screening across the wider family unit. When planning new (or refining existing) childhood screening programs, it is important to assess acceptability before implementation. This should include assessment of important issues such as information needs, timing of information, and when and where the screening should occur.

Introduction

Medical screening is a process whereby individuals undergo tests to determine whether they have, or have an increased risk of, a health condition. During childhood, there are many health conditions that can be screened for, including vision and hearing problems, heart defects, or biochemical genetic disorders. In 1968, Wilson and Jungner[1] defined criteria to be used to guide the selection of health conditions to be screened. Since then, there have been many advances in both diagnostic and therapeutic interventions. As such, a modified screening criterion was proposed

* Corresponding author. Tel.: +44 114 222 0799; fax: +44 114 222 0749.
E-mail addresses: j.carlton@sheffield.ac.uk (J. Carlton), h.griffiths@sheffield.ac.uk (H.J. Griffiths), a.m.horwood@reading.ac.uk (A.M. Horwood), P.P.Mazzone@sms.ed.ac.uk (P.P. Mazzone), rachel.walker@sheffield.ac.uk (R. Walker), simonsz@compuserve.com (H.J. Simonsz).
[e] Present address: Muir Maxwell Epilepsy Centre, Centre for Clinical Brain Sciences, University of Edinburgh. 20 Sylvan Pl, Edinburgh, EH9 1UW, UK.
[f] Present address: Faculty of Social Sciences, University of Sheffield, ICOSS, 219 Portobello, Sheffield, S1 4DP, UK.

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by Anderman et al.[2] The criteria 'The test should be acceptable to the population,'[1] and 'The overall benefits of screening should outweigh the harm'[2] relate to the acceptability of the screening program. Acceptability of healthcare interventions is a challenging construct. Sekhon et al.[3] acknowledged that there is little guidance on how to define acceptability. They defined acceptability to be 'a multifaceted construct that reflects the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on anticipated or experiential cognitive and emotional responses to the intervention.' They proposed a theoretical framework of acceptability. This includes affective attitude (how an individual feels about the intervention), burden (the perceived amount of effort that is required to participate in the intervention), ethicality (the extent to which the intervention has good fit with an individual's value system), intervention coherence (the extent to which the participant understands the intervention and how it works), opportunity costs (the extent to which benefits, profits, or values must be given up to engage in the intervention), perceived effectiveness (the extent to which the intervention is perceived as likely to achieve its purpose), and self-efficacy (the participant's confidence that he/she can perform the behavior(s) required to participate in the intervention). For childhood screening, there is further complexity as acceptability can be applied to both the individual (i.e., the child) and the caregiver (i.e., the parent or guardian).

Over recent years, there has been increasing demand on healthcare systems.[4,5] Population growth and life expectancy has increased, placing additional stress on existing healthcare systems.[6,7] Advancements in technologies to aid diagnosis and management of health conditions have also contributed to stretched resources.[8,9] Consequently, existing and proposed interventions are examined to ensure that they are both clinically effective and cost-effective.[10] However, the practical and ethical implications on families and children when screening services are planned or reviewed should also be considered.[11] To our knowledge, there has been no review that examines the acceptability of childhood screening interventions. The overall aim of this review was to assess the acceptability of childhood screening interventions with a view to identifying which factors to consider when planning or modifying childhood screening programs to maximize participation and uptake. We applied the framework outlined by Sekhon et al.[3] to establish which aspects of acceptability are most commonly evaluated and which research methodology is used.

Methods

An information specialist was consulted in developing the appropriate search strategy. One researcher (M.P.P.) conducted the searches. Search terms included in the review included the following:

i. Children (and derivatives)
ii. Screening (and derivatives)
iii. Acceptability terms

No restriction on the publication date was applied to the search strategy. Full details of the search strategy are provided in Appendix 1. The electronic databases searched for the systematic review were MEDLINE, EMBASE, PsycINFO via Ovid, CINAHL, and the Cochrane Library. All databases were searched from inception. Searches were conducted between May 1, 2018, and May 5, 2018. An updated search was performed in January 2020 to include publications from January 2018 to January 21, 2020. The following eligibility criteria was applied to the search results: published as a full-text original research article (i.e., not including abstracts, editorials, reviews, opinion pieces, or letters to the editor), inclusion of a postnatal screening program (i.e., not antenatal screening), child health condition screening programs (i.e., not adolescent and/or adult screening or the vaccination program), and child and/or parental perspectives (i.e., not healthcare worker perspectives). Studies that solely included healthcare worker perspectives were excluded. The protocol was registered with PROSPERO.[12]

To apply the eligibility criteria for the selection of articles from the search results, the following steps were performed: (1) two reviewers (M.P.P. and C.J.) undertook 'filtering of titles' independently. Where there was disagreement, articles were retained, and the abstract was scrutinized; (2) two reviewers (M.P.P. and C.J.) undertook 'filtering of abstracts' independently. Where there was disagreement, articles were retained, and the full text was scrutinized; and (3) 'filtering of full-texts' by three reviewers (M.P.P., C.J., and G.H.J.). Discussion and consensus had to be reached for an article to be included within the review.

Articles to be included in the review were assessed against the seven component constructs proposed by Sekhon et al.[3] by two reviewers (C.J. and G.H.J.). Any disagreement was resolved through discussion. Data were extracted by one reviewer (C.J.) using a piloted data collection form. Studies were examined to determine whether acceptability was assessed prospectively, concurrently, or retrospectively; categorized as to which acceptability construct was assessed; and categorized based on the study methodology. The type of childhood screening, country where screening occurred, and details of the study participants (child or parent/carer) were also noted.

Results

The database searches identified 4529 references. A total of 149 full-text articles were retrieved for further examination. From these, 103 articles were rejected as they failed to meet the inclusion criteria. A total of 46 publications are included in this review (see Fig. 1). The summary of findings for the included studies is shown in Table 1.

Of the 47 studies included in the review, most were conducted in the United States of America (USA) (n = 14), the United Kingdom (UK) (n = 12), the Netherlands (n = 4), Australia (n = 2), Canada (n = 2), and Sweden (n = 2) (Table 1). The majority of studies (55%) were published between 2010 and 2018. The content of the screening programs included is shown in Table 1. These were newborn blood spot screening (to identify biochemical and endocrine genetic disorders) (n = 22), neonatal hearing screening (n = 13), Duchenne muscular dystrophy (DMD) screening (n = 4), cystic fibrosis screening (n = 3), screening for congenital heart defects (n = 2), screening for congenital hypothyroidism (n = 1), and screening for hip dysplasia (n = 1). The details of which biochemical and endocrine genetic disorders were screened as part of newborn blood spot screening programs were not clearly reported, but the program typically included screening for conditions such as phenylketonuria and sickle cell disease, among others. Most of the studies (n = 44) concerned neonatal screening.

Acceptability was assessed quantitatively (n = 30), qualitatively (n = 26), and by a combination of methods (n = 10). Of the studies that adopted quantitative methods, the majority of studies used their own questions or questionnaire, or modified existing questionnaires. Some studies did include validated questionnaires, including the Beck Anxiety Inventory, State-Trait Anxiety Inventory questionnaire, Center for Epidemiological Studies Depression Scale, the depression scale of the Hospital Anxiety and Depression Scale, the Parenting Stress Index, and the General Health Questionnaire. Of the studies that adopted qualitative methods (n = 26), most involved interviews (n = 13) and seven studies undertook focus group sessions (n = 7). Some studies issued questionnaires that

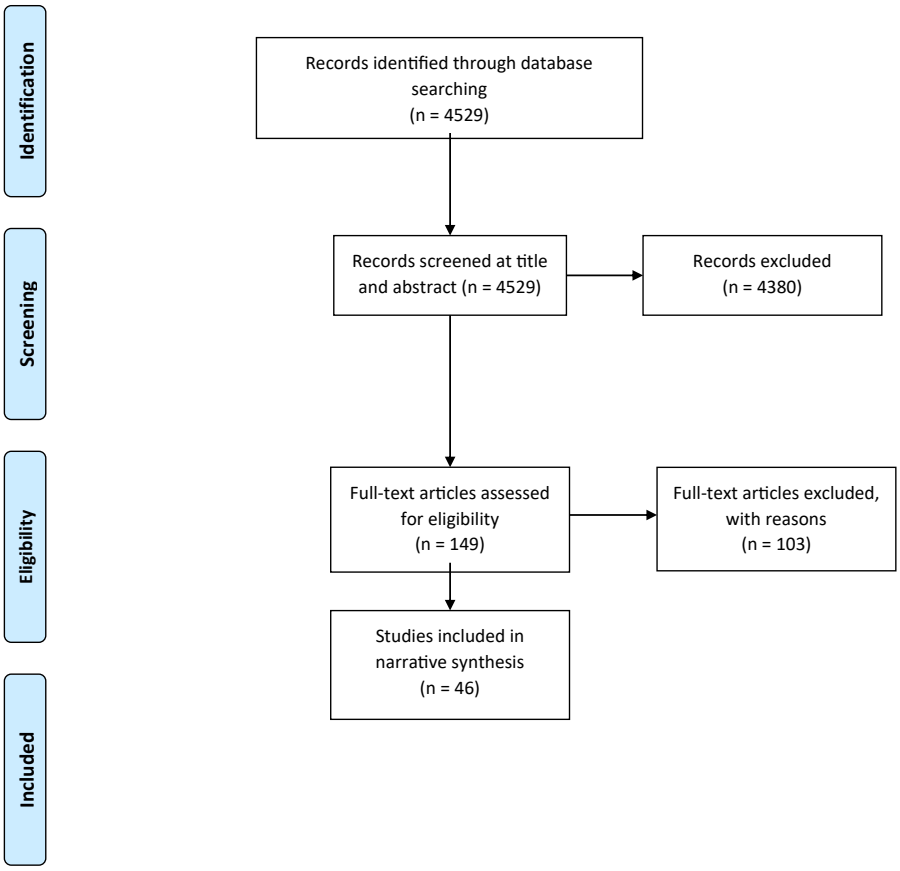

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2009 Flow Diagram: study identification.

incorporated some open-ended questions, and the free text was analyzed qualitatively (n = 8) (see Table 2).

Table 3 shows only three studies assessed acceptability at the time of screening.[13–15] The majority of studies assessed acceptability retrospectively (n = 40).[13,14,16–52] Ten studies assessed acceptability prospectively.[24,28,31,35,46,53–57] The majority of studies examined acceptability with respect to affective attitude (n = 41) and intervention coherence (n = 31). Other acceptability constructs assessed included burden (n = 9), ethicality (n = 5), perceived effectiveness (n = 9), opportunity costs (n = 6), and self-efficacy (n = 4). Most of the studies assessed more than one construct of acceptability. No study assessed all seven acceptability constructs (Fig. 2).

Affective attitude

In the context of screening, this is how a parent feels about the screening program itself. A total of 41 studies that assessed this concept were identified.[13–17,19,21,24–31,33–35,38–47,49–58] Most of the studies also included some form of assessment of parental beliefs on whether screening was thought to be of value.[31,39,41,58] Other studies also reported on parental satisfaction, specifically for screening service, be it in terms of receiving results or the screening test(s) not causing any discomfort to the parents' child.[17,19,33] Not all participants within the studies reported favorably. Tariq et al.[46] reported some parents (n = 10, 4%) to view the test for congenital hypothyroidism to be 'unimportant,' with some parents (n = 8, 3%) considering it to be a painful procedure for their child.

Burden

Nine studies explored the impact of burden.[16,30,33,34,40,43,52,54,57] The burden associated with screening varies from one screening program to another. The amount of effort required for the parent/caregiver to support the intervention (i.e., take the child for testing) can be considered as burdensome. When screening can occur in venues that required minimal effort from the parent (i.e., within the hospital or in the home), the acceptability of the screening is increased.[30,40,52] The burden of attending the appointments owing to work commitments or difficulties with transport can lead to non-attendance.[43] Financial burden may also be a factor as some parents reported their medical insurance did not cover the screening test(s).[16,34,43] Some studies inferred burden by parental observations of discomfort of testing on the child.[33]

Ethicality

Nine studies were categorized as assessing ethicality.[13,19,28,30,40–42,49,57] Some studies included assessments of beliefs with regard to the screening, including moral and religious views.[13,42,49] Parsons et al.[13] reported some mothers consented to screening for DMD as they approved of all screening. In a separate study, Parsons et al.[40] highlighted some mothers felt so positively toward newborn screening that they felt it should be made compulsory.

Intervention coherence

Thirty-one studies identified within this review investigated parental understanding of the screening program itself, and/or the potential consequences of a confirmed diagnosis of the target

Table 1
Summary of findings of the included studies.

Reference	Year	Country in which screening occurred	Condition	Age of the child subjected to screening	N (no. of mothers)	Aim	Summary
Akilan et al.[16]	2014	South India	Hearing	<2 years[a]	83 (83)	To review an existing rural community-based screening project	Community leaders played an important role in facilitating better coverage.
Al-Sulaiman et al.[17]	2015	Saudi Arabia	NBS	Newborn[a]	425 (425)	To assess the attitude and knowledge of mothers toward the NBS program	Positive attitude toward the NBS program; however, better communication is needed to increase awareness.
Araia et al.[18]	2012	Canada	NBS	Newborn, 24 −72 hrs after birth	750 (750)	To identify elements of NBS education and their associations with mothers' knowledge and satisfaction levels	Education and information before screening is important, particularly on the purpose, benefits, process, and possible results of screening.
Christie et al.[53]	2013	Australia	NBS for FXS	Newborn, 24 −72 hrs after birth	1971 (1971)	To determine feasibility and accuracy of two concurrent testing methodologies; to determine postnatal mothers' acceptance and attitudes to screening and reasons for accepting or declining participation; to assess the impact of diagnosis of a child with an abnormal result	Mothers considered an early diagnosis beneficial. Some were anxious about potential test results; others felt their feelings toward their newborn may change if he/she was diagnosed positive. High participation rates and maternal attitudes indicate a high level of maternal acceptance and support for screening.
Crockett et al.[19]	2005	UK	Hearing	Newborn OAE testing within 48 h of birth HVDT at 6–8 months of age	90 (90)	To compare the impact of two screening tests (newborn hearing screening − OAE test and HVDT) and screening recall on maternal anxiety and satisfaction	No significant differences were found (with respect to maternal anxiety, worry, and certainty) between the two tests.
Crockett et al.[20]	2006	UK	Hearing	Newborn within 48 h of birth	344 (344)	To describe the impact of newborn screening on maternal anxiety and to examine the impact of knowledge	Understanding the three screening recall systems may avoid some anxiety.
Cyrus et al.[21]	2012	USA	DMD	12 months	138 (120)	To assess the desirability of DMD screening, the effectiveness of the consent process, and the feasibility of screening in a pediatric office (i.e., after the newborn period)	Parents indicated broad support of screening. Parents understood the risks and benefits of screening. DMD screening is feasible in a pediatric office.
Danhauer and Johnson[22]	2006	USA	Hearing	Newborn[a]	36 (NR)	To assess parents' perceptions of an emerging community-based program in which screening and/or follow-up testing was provided on an 'outpatient' basis through a private practice	Parents were generally positive about all phases of screening. Findings were consistent with those reported from hospital-based programs.
Davis et al.[23]	2006	USA	NBS	Newborn[a]	51 (48)	To gather opinions about the content and timing of newborn screening education to inform recommendations	Parents had limited knowledge and awareness of NBS. Parents wanted concise information on all aspects of screening including benefits, need for retesting, and importance of follow-up (if required). Parents wanted verbal information from the provider and brochures. Parents felt information should be provided in the third trimester of pregnancy.
Detmar et al.[24]	2007	Netherlands	NBS	1st week of life	29 (22)	To investigate the preferences and views of parents and future parents with respect to information about, and consent to, neonatal screening and the possible expansion of the program	Parents were not well informed about what the test involves and viewed it as a routine procedure. If the program was to be expanded, parents would like to be informed earlier, preferably during pregnancy. Most parents preferred an opt-out consent approach.
Din et al.[54]	2011	USA	NBS CMV infection	Newborn[a]	3922 (NR)	To assess attitudes toward newborn screening for CMV	Among most parents, costs, worry, and anxiety associated with newborn screening for CMV would be acceptable. A minority of the parents weakly opposed to newborn screening for CMV.
Etchegary et al.[25]	2016	Canada	NBS	Newborn[a]	32 (30)	To explore parent and HCP experiences of NBS practices	Three themes were identified: offer of consent; content and timing of information; and importance of parental experiences for consent decisions. NBS was viewed as 'routine,' with little evidence of an informed consent process.

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Table 1 (*continued*)

Reference	Year	Country in which screening occurred	Condition	Age of the child subjected to screening	N (no. of mothers)	Aim	Summary
Fitzgerald et al.[55]	2017	Ireland	NBS	Newborn[a]	662 (662)	To determine if antenatal women received information about NBS in the antenatal period and to evaluate their knowledge and attitudes about NBS	All participants felt information should be given before birth. Information given about NBS in the antenatal period is inconsistent; consequently, awareness is limited. Mothers require information to be provided in a more structured format.
Hargreaves et al.[26]	2005	UK	NBS	Newborn[a]	47 (42)	To examine parents' and HCPs' views on informed choice in NBS and assess information and communication needs	Parents and HCPs recognize a tension between informed choice in NBS and PH screening in children. Clear, brief, and accurate parent information and effective communication between HCPs and parents, which take into account parents' information needs, are required for informed choice.
Hergils and Hergils[27]	2000	Sweden	Hearing	Newborn[a]	83 (NR)	To assess parental attitudes and concern of relation to universal NHS by OAE testing	Parents wanted early detection of hearing loss and the possibility of early intervention. Screening did not disturb the children. Most parents' experiences of NHS were positive and felt reassured by it.
Jatto et al.[56]	2018	Nigeria	Hearing	Newborn[a]	48 (48)	To determine the knowledge and perceptions of mothers of newborn children on hearing screening	Awareness of newborn screening was poor. Willingness to accept screening increased with increasing levels of education and increasing levels of socio-economic status. Knowledge of what factors are responsible for childhood hearing loss was poor.
Joseph et al.[28]	2016	USA	NBS	Newborn[a]	31 (31)	To examine the perspectives and values of diverse healthy pregnant women and parents of children diagnosed with a primary immunodeficiency disorder about traditional NBS and expanded NBS with the use of whole-genome sequencing.	Four themes emerged: (1) perspectives on traditional NBS, (2) informed consent, (3) return of results, and (4) storage and retrieval of results. Study participants desired greater inclusion in the NBS process. Parents voiced concerns about privacy and control over test results because of limited trust in the medical system and the state-run NBS program.
Khairi et al.[29]	2011	Malaysia	Hearing	Newborn[a]	78 (78)	To investigate maternal anxiety when the child had failed the test in the first stage of the UNHS	FP test results of the UNHS increased maternal anxiety.
Lam et al.[30]	2018	Hong Kong	Hearing	Newborn[a]	102 (102)	To investigate maternal knowledge, attitudes, and satisfaction of the UNHS	Information on the UNHS requires further details, particularly on implications of results and/or infant hearing development. Many did not understand the results.
Lang et al.[31]	2009	USA	NBS (CF and SCD)	Newborn	388 (388)	To examine maternal understanding of NBS for SCD and CF and their knowledge of the genetics, symptoms, and treatments of both conditions.	Poor understanding of NBS, greater familiarity with SCD, and significant knowledge gaps for both SCD and CF were found. There are many missed educational opportunities for educating parents about NBS and specific conditions included in NBS panels in both the obstetric clinics and the nursery.
Lipstein et al.[32]	2010	USA	NBS	Newborn[a]	45 (41)	To describe how parents consider disease and test characteristics while making decisions about newborn screening.	Parents' preferences differed based on experience with genetic conditions. Most parents wanted more detailed information. Some suggested optional testing. Understanding parents' decision-making processes and information needs would support development of screening policies that better address variations in preferences.

Magnuson and Hergils[33]	2009	Sweden	Hearing	Newborn (maximum 3 days postpartum)	49 (26)	To evaluate an existing newborn hearing screening program with regard to information and psychological support of parents	A majority of parents were in favor of screening, and screening caused little anxiety. Where more than one retest was required, parental anxiety increased and was linked to information needs.
Mak et al.[34]	2012	China	NBS	Newborn[a]	172 (NR)	To examine parental knowledge and attitudes toward the expanded NBS in Hong Kong	Parents favored having the expanded NBS in Hong Kong. Parental tolerance was high. Parents valued parental autonomy with informed consent and pretest counseling.
Moody and Choudhry[35]	2013	UK	NBS	Newborn <1 week	Survey, 140 (124) FG, 29 (27)	To explore perceptions and attitudes of parents and future parents to an expanded NBS in the UK and the necessary information provision and consent processes.	Parents want guaranteed information provision with clear decision-making powers and an awareness of the choices available to them. The difference between the existing NBS and expanded NBS was not considered to be significant enough by participants to warrant formal written, informed consent for expanded screening.
Narayen et al.[36]	2017	Netherlands	CHD	Newborn 1 hr after birth and at day 2/3	1172 (1172)	To assess the acceptability of PO screening to mothers after screening in the home setting	Overall, mothers were happy with the performance of the test, thought their baby was comfortable during screening, and did not feel stressed while the screening was performed. Most mothers would recommend PO and considered the test important.
Nicholls[37]	2012	UK	NBS	Newborn[a]	18 (16)	To explore whether parents experience the purported tension between compliance and achieving informed consent.	Two themes emerged relating to the voluntariness of choices: the expectation of compliance and presentation of information to promote compliance. In both cases, aspects of provision were noted as negatively impacting on the parents' perceived choice when accepting NBS.
Nicholls and Southern[38]	2013	UK	NBS	Newborn[a]	12 (10)	To understand the factors that influence parental decision-making in accepting NBS	Seven factors were identified: experience, attitudes to medicine, information-seeking behavior, perceived knowledge, attitudes to screening, perceived choice, and perceived decisional quality.
Parsons et al.[39]	2002	UK	DMD	Newborn[a]	97 (NR)	To evaluate the psychosocial implications of newborn screening for DMD	Most families with an affected boy identified through screening were in favor of NBS (to allow choice in future reproductive plans and time to prepare). Anxiety levels in the screened group were higher than in the control group, but were normalized during the study period.
Parsons et al.[58]	2005	Wales (UK)	DMD	Newborn[a]	1347 (NR)	To assess the effect of changing a protocol for DMD (to make the choice more explicit) as part of NBS.	The change in protocol resulted in increased satisfaction, awareness, and choice. No increase in worry was found, and parents indicated they felt a 'greater freedom' to refuse the test.
Parsons et al.[13]	2006	UK	DMD	Newborn[a]	1542 (1542)	To explore the reasons given by women for their decision about an optional newborn screening test for DMD.	Perceptions on screening were related to 3 overarching themes: screening as a routine procedure, screening for reassurance, and screening for disease detection.
Parsons et al.[40]	2007	Wales (UK)	NBS	Newborn[a]	18 (18)	To explore mothers' accounts of NBS and to explore the process of consent	Information about the screening was reported to be varied, and most mothers received this postpartum. Issues of consent were noted, and mothers felt that screening was perceived as routine.
Powell et al.[41]	2013	UK	CHD	Newborn[a]	813 (813)	To assess maternal acceptability of pulse oximetry screening for CHD in newborn infants and to identify factors predictive of participation in screening	Participants were mainly satisfied with screening. Anxiety of mothers given FP results was not significantly higher than of those given the TN result. Different ethnic groups had different participation readiness into the study,

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Table 1 (*continued*)

Reference	Year	Country in which screening occurred	Condition	Age of the child subjected to screening	N (no. of mothers)	Aim	Summary
							which may not reflect upon whether this would be observed in screening itself.
Quinlivan and Suriadi[42]	2006	Australia	NBS	Newborn[a]	200 (200)	To evaluate new mothers' opinions of genetics and newborn screening	Acceptance of screening is high, but mothers consider the need for consent to be mandatory.
Scheepers et al.[43]	2014	South Africa	Hearing	Newborn[a]	50 (NR)	To identify reasons why parents refuse newborn hearing screening and why some default on follow-up rescreening	Most frequent reasons for refusing screening were related to costs and knowledge about the screening process.
Skinner et al.[44]	2011	USA	NBS for FXS	Newborn[a]	1930 (1930)	To document rates of parental consent in a pilot study of screening for FXS, examine demographic characteristics of mothers who consented or declined, and describe the reasons for their decision.	A majority of parents accepted screening, but decision rates and reasons for accepting/declining varied in part as a function of race/ethnicity and in part as a function of what parents most valued or feared in their assessment of risks and benefits.
Stuart et al.[45]	2000	USA	Hearing	Newborn[a]	40 (40)	To determine whether mothers whose infants had failed NHS had more stress than those mothers whose infants had passed NHS	No significant difference was found between the two groups—those mothers whose infant had failed demonstrated equivalent stress levels as those mothers whose infants had passed.
Tariq et al.[46]	2018	Pakistan	Congenital hypothyroidism	Newborn[a]	355 (355)	To determine knowledge of congenital hypothyroidism and to assess the impact of health education on knowledge and attitudes toward screening	Most mothers were unaware of congenital hypothyroidism and its implications. Awareness increased after the intervention survey.
Tluczek et al.[14]	1992	USA	CF (as part of NBS)	Newborn[a]	104 (66, plus 28 responses from both parents)	To examine parental knowledge of (1) the screening program, (2) understanding of negative results, (3) effects of screening-related anxieties, and (4) the effects of FP results	Parents had gaps in knowledge about screening, misconceptions about test results, and high levels of anxiety.
Tluczek et al.[47]	2005	USA	CF (as part of NBS)	Newborn[a]	28 (25)	To investigate the psychosocial effects on parents of infants with abnormal results in CF NBS that uses genetic testing	Most parents experienced high levels of emotional distress waiting for the sweat-test appointment (diagnostic test). Parental uncertainty and emotional distress were influenced by prior knowledge of NBS, CF, their own carrier status, adjustment to having a new baby, and physicians' approach to parents.
Tluczek et al.[47]	2009	USA	CF (as part of NBS)	Newborn[a]	193 (100)	To learn how parents were informed about NBS and obtain their suggestions for improving the process of educating parents about NBS	Parents described much inconsistency in the timing of information and methods used to inform them about NBS. Parents recommended improving communication about NBS at multiple points. Parents suggested that providers take time to explain the purpose and importance of NBS, which diseases are included in testing, and when to expect results.
Ulph et al.[49]	2011	UK	NBS	Newborn[a]	37 (28)	To explore the origins and content of service users' prior knowledge of universal antenatal and newborn screening for hemoglobin disorders.	Families influenced participants' screening knowledge, decisions, and service use. Families were often participants' main source of support.
Vohr et al.[15]	2001	USA	Hearing	Newborn 1st screening before discharge Rescreen 2–8 weeks after discharge	307 (307) 1st screen 40 (40) rescreen	To identify and compare the prevalence and degree of maternal worry about NHS at the time of an initial NHS and rescreening	Maternal worry was greater at the rescreening cf. screening. Those who reported greater worry at the time of the screening were more likely to be socio-economically disadvantaged. Maternal knowledge of screening increased between the two time periods, but the degree of worry was unchanged.

Waisbren et al.[50]	2003	USA	NBS	Newborn[a]	407 (254)	To assess the impact on families of a FP screening result compared with a normal result in the expanded newborn screening program.	Expanded newborn screening may lead to improved health outcomes. FP screening results may place families at risk of increased stress and parent-child dysfunction.
Weichbold et al.[51]	2001	Austria	Hearing	Newborn[a]	90 (90)	To test the hypothesis that there is a positive association between information and positive attitude to NHS	Most mothers who were in favor of screening had a greater knowledge about the hearing test. This included being present at the test and being aware of the result of the test.
Weinreich et al.[57]	2012	Netherlands	NBS for Pompe disease	Newborn[a]	613 (not explicitly reported)	To measure support for neonatal screening for Pompe disease in the general public and to compare it to support among (parents of) patients with this condition	Results suggest a rather high level of support for newborn screening for Pompe disease, not only among those who have personal experience of the disease but also among the general population.
Witting et al.[52]	2013	Netherlands	Hip dysplasia	3 months	703 (NR)	To investigate whether psychosocial factors (attitude, subjective norm, self-efficacy, perceived susceptibility, perceived severity, perceived effectiveness) predicted parental participation in the screening.	Attitude, subjective norm, self-efficacy, perceived susceptibility, and perceived effectiveness predicted parental participation in the screening. Emphasizing positive aspects of the screening, highlighting the effectiveness, removing practical barriers, and being conscious of the influential role of HCPs on decision-making are areas to focus on while organizing the screening.

CF = cystic fibrosis; DMD = Duchenne muscular dystrophy; CHD = congenital heart defect; FP = false-positive; FXS = Fragile X syndrome; HCP = healthcare professional; HVDT = health visitor distraction test; NBS = newborn blood spot screening; NHS = neonatal hearing screening; NR = not reported; OAE = otoacoustic emission; PH = public health; PO = pulse oximetry; RHD = rheumatic heart disease; SCD = sickle cell disease; TN = true negative; UNHS = universal hearing screening program, UK = United Kingdom; USA = United States of America.

[a] Exact age not defined.

condition.[13,14,16–18,20–26,28–31,34,35,38,40,43,46–52,55,56,58] Studies examined the issues of parental knowledge, receipt of information, and previous experience of screening and experiences of friends and/or family members. Some studies explored issues of consent, which also included parents having sufficient information and appropriately timed information to allow for informed consent. Some parents recalled that newborn screening was offered as a choice where active consent was given, whereas other parents were less certain as to whether they did provide consent. Even within the same study cohort, parental accounts with regard to the issue of consent for screening varied.[25] For some parents, the screening process was 'routinized,' and that this can be inadvertently presented as compulsory.

Opportunity costs

Six articles identified issues with regard to opportunity costs.[16,23,26,34,43,54] Some studies discussed the consequences of direct financial costs on attending screening and whether such costs were covered by medical insurance.[23,43] Some parents were not concerned about the costs of testing, and others expressed a willingness to pay.[34,54] However, some parents stated that the expense of additional (screening) tests would result in the refusal of any advised additional testing.[23] One study reported that attendance to screening would come at a cost of missing work and giving up time with other children/family responsibilities.[16]

Perceived effectiveness

Perceived effectiveness was studied in nine studies.[14,16,28,32,38,39,41,43,52] Some studies reported that parents either had doubts in the effectiveness of the test, had doubts in the accuracy of results, or even had distrust of the healthcare system.[14,32,39,41,43,52] Some parents noted that screening would not be offered if it had not already been reviewed or assessed as being acceptable by experts, including medical professionals.[38]

Self-efficacy

Four studies were categorized as assessing for self-efficacy.[16,23,34,52] Parents reported that while they wanted information about the screening process, they noted that the timing the information was received was not appropriate. They felt overwhelmed with information and were 'often exhausted.'[23] The context of exhaustion may be particularly pertinent to screening programs that occur within the first few weeks/months of life. Generally, parents were confident that they were able to arrange other responsibilities to make time to attend for screening (and/or referral) appointments.[16,34,52]

Discussion

Acceptability of the childhood screening program is a relatively under-researched area. A key objective of this review was to identify factors to be considered to encourage participation in childhood screening programs, thereby maximizing the program's cost-effectiveness. Two of the most common constructs identified from the included studies were affective attitude (how the parent feels about the screening program) and intervention coherence (parental understanding of the screening program itself and/or the potential consequences of a confirmed diagnosis of the target condition). Determining how a parent or guardian feels about screening could be considered as an important first step when considering implementing new (or refining existing) childhood screening programs.[59,60] If parents' views are such that they feel

Table 2
Study methodologies of the included studies and types of data collection method(s).

Reference	Both	Quantitative assessment instruments											Qualitative methods		
		Own	EDS	STAI	IoE scale	CHQ-PF28	BAI	GHQ	HADS	PSI	CES-D	CST	Int.	FG	Free-text
Akilan et al.[16]	✗													✓	
Al-Sulaiman et al.[17]	✗	✓													
Araia et al.[18]	✗	✓													
Christie et al.[53]	✓	✓	✓	✓	✓										✓
Crockett et al.[19]	✓	✓		✓											
Crockett et al.[20]	✓	✓		✓											
Cyrus et al.[21]	✓	✓													✓
Danhauer and Johnson[22]	✓	✓													✓
Davis et al.[23]	✗												✓	✓	
Detmar et al.[24]	✗													✓	
Din et al.[54]	✗	✓													
Etchegary et al.[25]	✗												✓		
Fitzgerald et al.[55]	✗	✓													
Hargreaves et al.[26]	✗												✓	✓	
Hergils and Hergils[27]	✗														✓
Jatto et al.[56]	✗	✓													
Joseph et al.[28]	✗													✓	
Khairi et al.[29]	✗						✓								
Lam et al.[30]	✗	✓													
Lang et al.[31]	✗	✓													
Lipstein et al.[32]	✗													✓	
Magnuson and Hergils[33]	✗												✓		
Mak et al.[34]	✗	✓													
Moody and Choudhry[35]	✓	✓												✓	
Narayen et al.[36]	✗	✓													
Nicholls[37]	✗												✓		
Nicholls and Southern[38]	✗												✓		
Parsons et al.[39]	✓	✓		✓				✓					✓		
Parsons et al.[58]	✓	✓													✓
Parsons et al.[13]	✓	✓													✓
Parsons et al.[40]	✗												✓		
Powell et al.[41]	✓	✓		✓					✓						✓
Quinlivan and Suriadi[42]	✗	✓													
Scheepers et al.[43]	✗												✓		
Skinner et al.[44]	✗												✓		
Stuart et al.[45]	✗														
Tariq et al.[46]	✗	✓													
Tluczek et al.[14]	✗	✓													
Tluczek et al.[47]	✓										✓		✓		
Tluczek et al.[47]	✗												✓		
Ulph et al.[49]	✗												✓		
Vohr et al.[15]	✗	✓													
Waisbren et al.[50]	✗									✓		✓			
Weichbold et al.[51]	✗	✓													
Weinreich et al.[57]	✓	✓													✓
Witting et al.[52]	✗	✓													

Both = the study used both quantitative and qualitative methods; EDS = Edinburgh Depression Scale; STAI = State-Trait Anxiety Inventory; IoE scale = Impact of Event Scale; CHQ-PF28 = Child Health Questionnaire Parent Form 28 items; BAI = Beck Anxiety Inventory; HADS = Hospital Anxiety and Depression Scale; PSI = Parenting Stress Index; CES-D = Center for Epidemiological Studies Depression Scale; CST = client satisfaction tool; Int. = interview; FG = focus group; GHQ = General Health Questionnaire.

negatively about the screening program, this is likely to affect attendance and therefore efficiency of the program itself. Parental beliefs, understanding, and knowledge of the screening program (including what it entails and what the potential consequences may be) are influenced by information. The amount of information and timing of information is important not only to ensure parents understand the screening process but also to ensure that informed consent to participate in the screening program can be obtained.[23–25,35] It is therefore important to fully consider the information needs of parents while planning and implementing childhood screening programs.[13,20,23,25,29,32,38] Information needs may differ between groups and populations.[11] A standardized approach across a whole country may not be appropriate, and localized documents (or other information resources) should be considered. Other issues identified included the burden of the screening program and any costs associated with the screening program.[16,23,26,34,43,54] Acceptability was noted to be influenced by minimal effort in participating in the screening process (i.e., whether the screening was undertaken at a convenient location, such as within the hospital or in their own home [30,40,52] and whether the costs were minimized). When screening exists as part of a suite of health checks, this makes the screening more acceptable to parents.[11] All of these factors may influence screening uptake and attendance. Not all costs were noted to be direct financial costs (such as paying for the screening tests), but could also be related to taking time to travel to the venue where the screening is carried out, how long the screening may take, and any loss of income due to taking time off work.[16] It was difficult to draw any firm conclusions on whether potential financial implications of attending screening could influence the acceptability of such programs. The studies identified varied in the country setting, from low-income countries (India)[16] to upper middle-income countries (China and South Africa)[43,54] to high-income countries (UK and USA).[23,26] Consideration of how healthcare systems are funded is important, particularly if parents

Table 3
Assessment of acceptability and constructs included.

Reference	When acceptability was assessed			Component constructs of acceptability						
	Prospective	Concurrent	Retrospective	Affective attitude	Burden	Ethicality	Intervention coherence	Opportunity costs	Perceived effectiveness	Self-efficacy
Akilan et al.[16]			✓	✓	✓		✓	✓	✓	✓
Al-Sulaiman et al.[17]			✓	✓			✓			
Araia et al.[18]			✓				✓			
Christie et al.[53]	✓			✓						
Crockett et al.[19]			✓	✓		✓				
Crockett et al.[20]			✓				✓			
Cyrus et al.[21]			✓	✓			✓			
Danhauer and Johnson[22]			✓				✓			
Davis et al.[23]			✓				✓	✓		✓
Detmar et al.[24]	✓		✓	✓			✓			
Din et al.[54]	✓			✓	✓			✓		
Etchegary et al.[25]			✓	✓			✓			
Fitzgerald et al.[55]	✓			✓			✓			
Hargreaves et al.[26]			✓	✓			✓	✓		
Hergils and Hergils[27]			✓	✓						
Jatto et al.[56]	✓			✓			✓			
Joseph et al.[28]	✓		✓	✓		✓	✓		✓	
Khairi et al.[29]			✓	✓			✓			
Lam et al.[30]			✓	✓	✓	✓	✓			
Lang et al.[31]	✓		✓	✓			✓			
Lipstein et al.[32]			✓	✓					✓	
Magnuson and Hergils[33]			✓	✓	✓					
Mak et al.[34]			✓	✓	✓		✓	✓		✓
Moody and Choudhry[35]	✓		✓	✓			✓			
Narayen et al.[36]			✓	✓						
Nicholls[37]			✓	✓						
Nicholls and Southern[38]			✓	✓			✓		✓	
Parsons et al.[39]			✓	✓					✓	
Parsons et al.[58]			✓	✓			✓			
Parsons et al.[13]		✓		✓		✓	✓			
Parsons et al.[40]			✓	✓	✓	✓	✓			
Powell et al.[41]			✓	✓		✓			✓	
Quinlivan and Suriadi[42]			✓	✓		✓				
Scheepers et al.[43]			✓	✓	✓		✓	✓	✓	
Skinner et al.[44]			✓	✓						
Stuart et al.[45]			✓	✓						
Tariq et al.[46]	✓		✓	✓			✓			
Tluczek et al.[14]		✓	✓	✓			✓		✓	
Tluczek et al.[47]			✓	✓			✓			
Tluczek et al.[47]			✓				✓			
Ulph et al.[49]			✓	✓		✓	✓			
Vohr et al.[15]		✓		✓						
Waisbren et al.[50]			✓	✓			✓			
Weichbold et al.[51]			✓	✓			✓			
Weinreich et al.[57]	✓			✓	✓	✓				
Witting et al.[52]			✓	✓	✓		✓		✓	✓

are meeting the financial costs of screening. Studies have shown that socio-economic status and risk of having disease (or health condition) do influence screening participation.[11,61–63] Although it can be hypothesized that parents with lower socio-economic status may find screening less acceptable, the studies identified in this review can neither support nor refute this hypothesis. Further studies are required to fully understand the financial burden of screening, either the cost of testing and/or the costs incurred owing to attending screening (such as travel costs and lost income).

The studies identified in this review were conducted on small populations. All the studies assessed acceptability from a parental (often maternal) perspective rather than from the individual's perspective. None of the studies explicitly explored whether acceptability differed within different population groups (such as ethnicity, educational status, and so on). Further research is required to investigate the acceptability of childhood screening programs across the wider family unit, with increased inclusion of modern-day parenting situations and roles.

A mixture of study methodologies was used to assess childhood screening acceptability. For the studies that used quantitative methods, existing validated questionnaires were administered to parents to measure anxiety associated with the screening process. In these studies, the level of anxiety was used as a proxy for acceptability.[19,20,29,39,41,53] However, it must be acknowledged that anxiety is in itself a multifaceted construct. Many parents find having a new baby a stressful time, even when good support

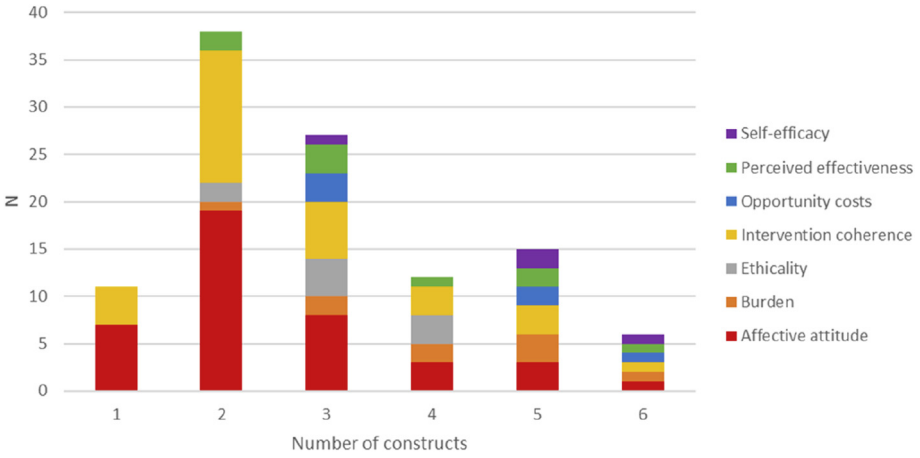

Fig. 2. Number of acceptability constructs reported within identified studies.

networks (such as friends and family) are in place. Increased levels of anxiety with regard to the time of neonatal screening may occur irrespective of whether a parent has anxiety with regard to the screening itself. Therefore, quantifying anxiety using an existing questionnaire(s) may not be the most appropriate method to understand what impact childhood screening has on a parent. To fully understand the individual's behaviors and feelings, qualitative research methods are required. The use of qualitative research methods facilitates an in-depth understanding of behavior and the reasons that govern that behavior[64] and may provide deeper insight into how parents feel about childhood screening programs.

The majority of the studies identified in the review were retrospective in nature, and the results perhaps should be treated with caution. The parental perspective of acceptability may have been influenced by the outcome of the screening program itself, that is, whether the child is found to have (or not) the condition for which the child was screened. This factor was not always disclosed in the included studies. Some may argue that acceptability is linked to satisfaction; however, Sekhon et al.[3] state there is a difference between these concepts. They argue that satisfaction can only be assessed retrospectively, whereas acceptability can be assessed both retrospectively and prospectively. Another important issue relating to retrospective assessment of acceptability is the timing of assessment in relation to the screening episode, i.e., how 'retrospective.' Recall bias is an important consideration when interpreting the results of any study.[65,66] Future studies will need to determine whether any impact of acceptability is present only in the short term (i.e., soon after the screening intervention) or more in the long term (i.e., months or even 1 year after screening). For example, issues of exhaustion and poor timing and information overload[16,23,34,52] may only be apparent or measurable if acceptability is assessed in the short term.

Limitations

This review is not without its limitations. Owing to the limited number of studies identified, no assessment (and therefore restriction) of study quality was performed. It is possible that bias exists within the studies, and the conclusions of individual study findings should be considered against issues such as design bias, sampling bias, measurement bias, interview bias, response bias, and reporting bias. The varied study outcomes and methodologies meant that meta-analysis and synthesis beyond a narrative review was not possible. A further limitation is that of acceptability construct categorization. Some of the constructs within the framework are linked, for example, burden and opportunity costs. The burden of attending a screening program may include time (which may include time off work, which could incur a cost), travel (which will incur a cost), and psychological burden (such as anxiety or worry). Affective attitude and perceived effectiveness are also related. Both constructs are associated with parental knowledge and understanding and information needs. Intervention coherence may relate to parental understanding of what the screening test(s) involves, any risks associated with the test(s), the consequence of a 'positive' screening result, consent for the screening test(s) to take place, and the effect involved in consenting to the screening program. Perceived effectiveness of screening centers on how well/accurate the screening test(s) is in being able to provide an indication of whether a child has the target condition, i.e., is the screening going to work? Similarly, affective attitude and ethicality are also linked. Although studies were assessed by two reviewers, there were inconsistencies with categorizations. Disagreements occurred when the results of the included studies could infer assessment of a construct (i.e., parental feelings of the screening program could infer the ethicality of screening). Most studies concerned neonatal screening. The findings may not apply to screening in older children. The acceptability of screening in older children may include other constructs, and the perspective of the child could also be considered.

Conclusions

Acceptability of childhood screening programs is an under-researched area. The aim of the review was to assess the acceptability of childhood screening interventions with a view to identify which factors to consider when planning or modifying childhood screening programs to maximize participation and uptake. We identified that in the context of childhood screening programs, acceptability was often determined by assessing parental knowledge and understanding of the screening process, the testing procedure(s), and consent. The emotional impact of childhood screening explored maternal anxiety levels associated with the timing of the screening process and the impact of any false referral. There are evidence gaps, and further studies are required to examine the acceptability of childhood screening across the wider family unit, including the child themselves (for screening in older children). While planning new (or refining existing) childhood screening programs, it is important to assess acceptability before

any implementation. The results of such studies can then inform and address issues such as information needs, timing of information, and when and where the screening should occur.

Author statements

Ethical approval

Not applicable.

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Competing interests

The authors have nothing to declare.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.puhe.2021.02.005.

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Themed Paper — Short Communication

A funfair without the candy floss: engaging communities to prevent diabetes in Nepal

J. Morrison [a, *], A. Arjyal [b]

[a] University College London, Institute of Global Health, London, UK
[b] Herd International, Thapathali, Kathmandu, Nepal

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ABSTRACT

Objectives: The World Health Organization estimates that 422 million people have diabetes, three-quarters of whom live in low- and middle-income countries. Global action plans to address non-communicable diseases (NCDs) recognise the centrality of community engagement to create an enabling environment within which to address risk factors.
Study design: In this article, we describe and critically reflect on a cocreated community engagement approach to address type 2 diabetes in the southern plains of Nepal. We coproduced the engagement approach with 40 artists from the Janakpur Women's Development Centre to create an environment for dialogue about diabetes and NCD risk between artists and the general public.
Methods: We used participatory action research to produce contextually relevant interactive methods and materials. Methods included artists' peer research to inform creative workshops, a drama performed in 19 villages and a two-day funfair in a public park. We used qualitative and participatory methods to analyse the effect of this engagement and reflect on lessons learned.
Results: Around 2000 people saw the drama, and around 4000 people attended the funfair. Community dialogue about prevention of diabetes was facilitated by drama and through games and songs at the funfair. Artists grew confident to interact with their peers and drama audiences about the causes of diabetes and prevention strategies. Despite crowds at the funfair, it was difficult to reach women because the venue was often used by men and boys, and patriarchal norms prevent women from free movement. Village interactions were able to engage a more mixed audience.
Conclusion: Innovative, asset-based community engagement about diabetes and other NCDs at scale is possible through locating, building on and strengthening community resources to address local health issues. Engagement could be enhanced by considering the gendered nature of community engagement spaces and by increasing opportunities for interaction between artists and the general public through more intimate and large-scale events.

Three-quarters of the 422 million people who have diabetes live in low- and middle-income countries.[1] In South Asia alone, an estimated 96 million people have diabetes. Ninety percent of these people have type 2 diabetes, a largely preventable disease.[2] There is an urgent need for multisectoral approaches to create enabling environments to address risk factors and ensure that community engagement is well integrated in non-communicable disease policies and plans.[3] Many multisectoral action plans approach community engagement through mass media,[4] despite limited evidence of its effectiveness in changing behaviours.[5] Although it may create an enabling environment, mass media provides inadequate opportunity for interaction to develop social support networks, which have been key to the success of peer and group-based interventions among high-risk populations.[6] Research from rural Bangladesh found that a participatory community group-based intervention reduced the combined prevalence of type 2 diabetes and hyperglycaemia by 21% in intervention areas when compared with control areas.[7] The interactive nature of the intervention was integral to its success.[8] There is a need to further innovate and develop effective community engagement interventions to address diabetes. We describe such an intervention in Nepal and reflect on lessons learned to inform future initiatives.

* Corresponding author. Tel.: +44 207 905 2303.
E-mail address: joanna.morrison@ucl.ac.uk (J. Morrison).

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We worked with 40 artists from the Janakpur Women's Development Centre in the southern plains of Nepal to cocreate a population-based community engagement approach for diabetes prevention. We sought to build on local assets of an established female community art centre and a strong tradition of Mithila art to engage communities about diabetes. Mithila art is traditionally painted on the outside of houses by women. A diverse and resilient group of artists work at the centre, many of whom have been socially and economically marginalised by illiteracy, unstable home environments, chronic illness, disability and widowhood. An asset-based approach seeks to use and build on local collective skills, resources, talents and relationships to improve health and well-being.

We used participatory action research,[9] collaboratively and iteratively designed the engagement based on context-specific artistic forms of expression and focused on local issues. To enable this, artists undertook 16 peer interviews to explore local experiences and understandings of diabetes. Initially, artists lacked confidence, and we developed this through training, practice, positive reinforcement and participatory development of visual tools (https://www.ucl.ac.uk/global-health/sites/global-health/files/pictorial_consent_process_final.pdf). We found that diabetes was poorly understood and beliefs about it being a communicable disease caused stress and anxiety among those with diabetes and their families, perpetuating social stigma. The cost of diabetes care was prohibitively high, which often caused guilt about the impact of diabetes on the family. Given the lack of specialist health care outside the capital Kathmandu, we agreed that a focus on prevention was a priority for engagement. A local health worker helped us to disentangle truths and untruths, and we designed art and games for a two-day funfair and created a travelling drama to promote active learning and stimulate conversations about diabetes (Fig. 1) (https://www.youtube.com/watch?v=8orIX40-ILw). Artists performed the drama in 19 villages and markets with audiences of around 100 people and hosted a two-day funfair in a public park in the urban town of Janakpur. Around 4000 people attended the funfair, 800 of whom had free blood glucose testing. We evaluated our community engagement through three focus group discussions (FGDs) with artists, two FGDs with women who had attended the funfair, one FGD with men who had attended and six artist peer interviews with those who had attended the funfair. Artists and researchers also engaged in critical self-reflection, making notes about what worked well and what worked less well throughout the engagement process.

At the funfair, giant snakes and ladders, games to knock over 'unhealthy' objects and games to feed 'unhealthy' figures 'healthy' food were popular among men and children, and women preferred the Zumba exercise routine. Stage shows of the drama and songs were more popular than the immersion tunnel and head-in-the-hole photo stands. Artists' confidence grew throughout the project, and they felt able to teach others about diabetes and to perform on stage. The travelling drama attracted large crowds in market areas, with many more men than in rural villages, where audiences were smaller and with a mix of men, women and children. Village dramas engaged the old and young, and the smaller audiences made it easier to initiate dialogue after the drama. Some community drama audiences felt that artists were acting inappropriately and feared their behaviour would erode social norms. An artist told us, "People were saying: 'These women have come to destroy our village.'" But as a group, their fear of social shame was less, and interactions with crowds afterwards led to increased understanding about why the women were performing and were effective in promoting dialogue about how to prevent and control diabetes.

Despite crowds at the funfair, we found it more difficult to reach women because the venue was not a place where women or families would usually go. Gender norms are patriarchal, and it is not considered decent for women to be outside of the home without purpose. Our mixed gender advisory committee recommended the funfair location—a large public park where men and boys often play cricket—but without adequate consideration of the gendered nature of the space. Conventional funfairs move from place to place. Taking the drama and the funfair around villages and more intimate locations such as schools or hospitals could enable equal access to participate among men and women and facilitate more interaction between artists and the public.

Innovative, asset-based community engagement at scale is possible through locating and strengthening community resources to address local health issues. Asset-based approaches have been critiqued for seeking to shift the focus away from structural inequalities driving poor health,[10] and we acknowledge that

Fig. 1. Artists perform a drama about how to prevent diabetes.

combining community engagement with health system strengthening to deal with the increased demand for services is important for such initiatives.

Finally, evaluating community engagement is challenging. Some have advised the capture of contribution as opposed to seeking causal attribution, whereas others have suggested realist approaches to consider complexity.[11] If the engagement is long-lasting and embedded, measurement of behavioural and biological outcomes is possible, for example, through cluster randomised controlled trial designs, wherein communities are randomly allocated to participate in the engagement process or to serve as control communities. Knowledge, practice and health outcomes such as prevalence of type 2 diabetes and intermediate hyperglycaemia could be measured and compared between trial arms.[7] But it is important to develop a robust theory of change to maximise the chance of success of the intervention. It is also important to take a pluralist approach to evaluation, using methods that acknowledge how success is viewed by the diversity of implementers and participants.[12]

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Ethical approval

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Competing interests

The authors have no competing interests to declare.

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Themed Paper – Short Communication

Art boxes supporting parents and infants to share creative interactions at home: an art-based response to improve well-being during COVID-19 restrictions

V.G. Armstrong*, J. Ross

University of Dundee, UK

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ABSTRACT

Objectives: This article seeks to demonstrate the impact of distributing boxes of art resources and guided activities for vulnerable parents and infants to do together at home.
Study design: Designed in conjunction with the local arts centre and the psychology team at the University of Dundee, the art boxes were a response to planned face-to-face art interventions with families being cancelled due to COVID-19 restrictions. The aim of the art boxes is to encourage parents to make art together with their infants, fostering connection through playful, creative shared experiences. This research is currently being expanded to reach out to new families through referrals from health visitors, family nurses, and charity partners.
Methods: Data is being collected on how the art boxes are experienced by families using a mixed-methods approach. Families complete feedback cards (online, or using the stamped addressed card included in the box) rating their experience on quantitative scales and providing open comments. Visual data are gathered through parents sharing images with us on social media. An initial sample of 10 participants has been interviewed using semistructured interviews, allowing more in-depth qualitative understanding of their experiences. These preliminary findings are discussed here.
Results: The thematic analysis of initial interviews provided a rich picture of the disconnection families experienced during lockdown, why art boxes may be beneficial to parental well-being, and the mechanisms by which the boxes may help to develop connections for the parent and infant together.
Conclusions: Preliminary findings show parents reporting feeling more confident and undertaking new activities which they plan to continue. This was of particular importance during lockdown where parents report opportunities for different experiences being more limited. Parent's describe positive playful interactions and reported improvements to their own well-being from doing creative activities together with their child. Analysis of these initial interviews gives a framework of barriers and supports to connection which highlights how art boxes can facilitate connectedness between dyads with the potential to strengthen attachments.

There is a developing case for the social benefits of art,[1] including the impact of arts on mental health and on the well-being of children. There are positive evaluations of participative arts in the early years[2] and research shows dyadic art therapy sessions can improve parental well-being and children's attachments.[3] However, we know that social factors impact upon arts participation[4] and in the light of the pandemic existing inequalities have been exacerbated,[5] as well as parental mental health difficulties).[6]

The Art at the Start project, a collaboration between University of Dundee and Dundee Contemporary Arts, has been developing a family programme of early years participative arts activities, including art therapy sessions targeting families of infants aged 0–3 years. These art therapy sessions take referrals from health and voluntary sector agencies, based on concerns that parents are vulnerable to low well-being and mental ill-health and infants may be at risk of attachment difficulties. When COVID-19 restrictions came into place, several planned art therapy groups were cancelled,

* Corresponding author. Psychology Department, University of Dundee, Nethergate, Dundee, DD1 4HN, UK. Fax: +1 07877 461 169.
E-mail address: V.G.Armstrong@dundee.ac.uk (V.G. Armstrong).

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affecting more than 40 families. We were concerned about the withdrawal of support for those families and their likely lack of resources to participate in some of the online activities being offered by ourselves and others over this time. To address this concern, we developed an innovative idea to maintain the families' engagement in joint art making. The art therapist produced boxes containing everything families needed to participate in 12 suggested activities (with variation for age and stage), including information on why these kinds of activities are beneficial. Boxes were sent during the highest restrictions in May 2020, by courier to avoid unnecessary risk from contact. Although these were not a replacement for art therapy, the activities were informed by what we have learnt in art therapy sessions about maximising connections. Activities were designed to encourage parents to make art together at home with their infants, fostering connection through playful, creative shared experiences(Fig. 1). With positive feedback from partner agencies and ongoing need, we were funded to roll out this scheme to reach at least 100 more families who we had not previously been involved with, between June and September when many restrictions remained. We have taken a mixed methods approach to collecting data including postcards with scaling questions in every box and gathering visual data by inviting families to share images through social media. This report focuses on our initial findings from semistructured interviews, conducted by phone, with a sample ($n = 10$) of parents involved, all of whom had their boxes for two months or more to allow time to take part in the activities. Our data provide promising insights into the mechanism by which this scheme may benefit parent-infant relationships.

Themes from interviews

Semistructured interviewing was chosen to give consistency whilst allowing some freedom to ensure that parents had understood the questions or to ask for further clarification or detail. Thematic analysis was undertaken on the transcripts of interviews using a reflexive model of practice.[7] In our analysis, we found a framework around barriers and supports to connectedness was a useful way of meaningfully describing all the data. A summary of our overarching themes and subthemes can be seen in Table 1.

Disconnecting experiences

All parents reported finding the lockdown period hard and it was clear this had impacted upon their well-being. Parents described the challenge of keeping occupied at home without their normal groups and social activities and the unavailability of shops. Some mentioned the challenge of occupying different ages of children. They reported feeling bored themselves alongside guilt about their infant's experiences. Parents also made statements that specifically reflected loneliness and isolation over this time. In particular, several mentioned not having 'mum friends' as they had not had any opportunity to make them. Connected to that feeling of isolation, a couple mentioned how different they felt their experiences had been to their peer groups without children. All parents reported the lack of supports available to them, with the withdrawal of parent support groups and home visits.

Fig. 1. A mother and infant exploring the contents of their art box together.

Qualities of art materials and boxes to support connected experiences

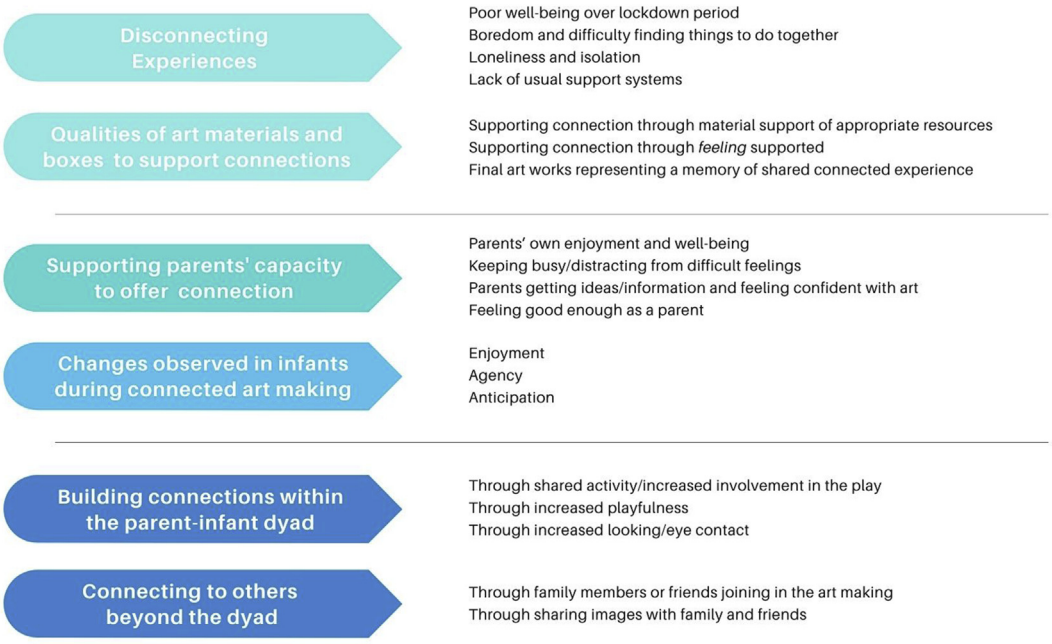
All parents reported appreciating the resources and that they would not have had them available without the packs, with reasons including unavailability of shops, not knowing where to go or what would be safe for their child. Having a resource which is age appropriate and available is important if we are to facilitate this kind of play. We asked about activity preferences and responses were broad with the largest number choosing paint. A particular point that was emphasised by the parents was the tactile quality of the materials and how these were appreciated by their infants. We noticed a repeated description from parents of the experience of actually opening up the box itself and exploring the contents with their infants. Other statements framed the art box as a gift. We think this represents the fact that the art boxes become a physical symbol that someone is offering support and that they are 'held in mind'. One parent made this explicit saying that they felt 'blessed' and that they were thought of. The final way in which the physical qualities of art making seemed to support connection was by providing a concrete reminder of positive moments. Parents talked about having art works up on display and of particular things that they would keep, such as clay handprints.

Supporting parents' capacity to offer connected experiences

To offer positive connected moments to their child a parent needs to have sufficient emotional availability. We found a number of themes about ways boxes supported the parents so that they in turn could connect to their infants. Parents reported positive feelings themselves from doing activities, including increased happiness, calm, fun and relaxation. Several mentioned their own enjoyment of the art making and others that the boxes had arrived at a good time when they were low. Parents appreciating having something planned that would fill their day. They reported liking to have a focus and that they felt less bored. Some also described how this was able to take their mind off difficulties. A theme emerged of parents feeling guilty about what they were doing with their infants and the art activities helping them to feel they were doing a good enough job.

There was also a practical aspect of parents getting ideas about the kinds of activities they could do and a prompt to do them. They all felt they had needed the instruction booklet, particularly for guidance around what works for younger ages and knowing what was safe. All parents reported feeling more confident from using the box and several described ways in which they were now able to adapt activities to suit or come up with ideas. Previously only three of the parents reported a limited amount of art making but following the box all stated that they would keep making art, some having already accessed online resources from the gallery.

Table 1
Changes to connection through shared art making at home.



Changes observed in the infant during connected art making

We asked about changes that parents had seen in their infants and identified three themes reflecting psychological changes for the infant. Parents described signs of enjoyment, that infants were happy, having fun and livelier. Parents also described behaviours that we would consider signs of agency – infants enjoying the consequences of their actions with the materials. For example, moving paint about on the paper, making choices or even getting art materials out for themselves. Parents also thought infants knew what was coming and were looking forward to it. This shows that infants were anticipating the activity, and it held positive associations for them.

Building connectedness within the dyad

All the parents reported feeling connected to their infant during the art activities and gave interesting insights about what aspects of the art making process might be supporting this connection. All talked about how the art activity was something shared, even that it was something that was meant to be done jointly. They reported being more involved, physically and emotionally, than when doing an equivalent activity, such as playing with toys. An increased amount of participation in play from parents will increase the opportunity for the dyad to have moments of positive connection. Parents also described being more playful with their infant during the art, similarly increasing potential for positive connected experiences. The strongest evidence for psychological connection was premised on eye contact, with parents describing their infants turning to look at them during art making, showing them materials and seeking eye contact. Parents reported responding to these connection seeking opportunities and their own positive feelings arising from them.

Connecting to others beyond the dyad

A final theme, surprising to us, was how the art boxes had helped the parent-infant dyads to connect with others. Half described family members or friends joining in with their art activity and that this had been a positive thing to share, even decreasing difficult sibling interactions. While not all parents had people around to become involved, they all brought up other ways in which they had shared art works with other people, either by giving physical artworks as gifts or by sharing images over social media. This seemed to have been particularly valued, perhaps because separated from those connections over lockdown.

Discussion

It seems clear that families used and valued the art boxes and increased their involvement in shared creative activities. Most interesting to us, is the insight into potential mechanisms by which the art boxes might be able to facilitate positive connected experiences for infants and their caregivers. The boxes supported parents so that their own well-being improved, potentially making them more available to their infant,[8] as well as giving those parents a feeling that they were doing a 'good enough' job for their infant and some relief that they had activity planned in their day. The process of art making itself seemed to promote connected experiences by encouraging playful, shared engagement in the activity. This was evidenced in the observation of increased eye contact. These kinds of connected interpersonal experiences between infant and caregiver are what help to build positive attachments[9,10] so could offer broad benefits to these families. We do not suggest that these packs could provide the equivalent level of benefit to a face-to-face service such as parent-infant art therapy[3] without the additional benefits of the therapists support for interactions and containment of difficult emotions. However, in the current circumstances these kinds of interventions could offer a useful resource to improve wellbeing during social distancing measures, where families do not have access to their usual activities, resources and social supports. As social distancing requirements are gradually relaxed, and public spaces reopen, this model of boxes may offer a potential mechanism to connect

families to the arts centre, who may not typically engage, and encourage them to participate in this community asset offering free, creative, family activity. Our study will continue to interview participants using these preliminary themes as a model, as well as analysing the data from quantitative feedback and using visual data in the form of images from parents to add insight about those aspects which they found important.

Author statements

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Competing interests

There are no competing interests to declare.

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Corrigendum

Corrigendum to "Maternal opioid use disorder at delivery hospitalization in a rural state: Maine, 2009–2018" [Public Health 181C (2020) 171–179]

S.M.B. Gabrielson [a], J.L. Carwile [b], A.B. O'Connor [c], K.A. Ahrens [a, *]

[a] Muskie School of Public Service, University of Southern Maine, Portland, ME, USA
[b] Division of Applied Health Care Delivery Science, Department of Medicine, Maine Medical Center, Portland, ME, USA
[c] Maine-Dartmouth Family Medicine Residency, MaineGeneral Medical Center, Waterville, ME, USA

The authors regret that the list of ICD-9 diagnosis codes used to capture maternal co-occurring conditions equivalent to ICD-10 diagnosis codes was incorrect. Correction of these errors does not change the overall findings or conclusions of the article, but does change the prevalence estimates for maternal co-occurring conditions shown in Table 1, Table 3, Fig. 2, the abstract, and several sections of the manuscript text.

In the abstract, the results should read as follows:

'The following conditions were more prevalent among women with OUD at delivery: hepatitis C, PR = 43.4 [95% CI: 37.5, 47.9]; other drug abuse or dependence, PR = 17.1 [13.8, 21.2]; alcohol abuse and dependence, PR = 8.4 [5.8, 12.3]; nicotine use, PR = 6.0 [5.9, 6.2]; cannabis use, PR = 5.7 [5.2, 6.2]; anxiety, PR = 2.7 [2.5, 2.9]; and depression, PR = 2.4 [2.2, 2.7].'

In the results, the fourth paragraph should read as follows:

'Other substance use disorders, certain mental health diagnoses, and hepatitis C diagnoses were more common among deliveries to women with OUD compared to women without OUD (Table 3). The co-occurring conditions with the largest difference in prevalence between deliveries with and without maternal OUD were hepatitis C (14.9% vs 0.4%, PR = 43.4 [95% CI: 37.5, 47.9]); other drug abuse or dependence (not including opioid or alcohol) (3.2% vs 0.2%, PR = 17.1 [13.8, 21.2]); alcohol abuse or dependence (0.8% vs 0.1%, PR = 8.4 [95% CI: 5.8, 12.3]); and nicotine use (63.5% vs 10.5%, PR = 6.0 [95% CI: 5.9, 6.2]). We observed an increase in hepatitis C, anxiety, cannabis use, and major depression at delivery hospitalization among women with OUD over the study period, which included the transition from ICD-9 to ICD-10 (October 1, 2015); a more moderate increase in non opioid substance use disorder was observed starting in 2016 (Fig. 2A). Among deliveries without OUD, a smaller increase in the diagnosis of anxiety, major depression, and cannabis use was observed over the study period (Fig. 2B). Nicotine use remained fairly steady over time in both groups.'

In the discussion, the fourth paragraph should read as follows:

'Our findings of co-occurring maternal conditions among women with OUD are generally consistent with the literature. An analysis based on national hospital discharge data from 1998 to 2011 found lower prevalence estimates for depression (8%) and anxiety (4%), and similar estimate for alcohol abuse or dependence (2%) among deliveries with OUD.[7] We found much lower prevalence of non-opioid drug abuse or dependence (3% vs. 22%)[7]; however, our definition of this condition was more limited (i.e. excluded cannabis and alcohol use). We found a low prevalence of alcohol abuse/dependence at delivery hospitalization, which concurs with prior literature suggesting alcohol use is unlikely to be diagnosed unless it is heavy use.[10] Further, estimating concurrent alcohol use among women with OUD is challenging as it is often not detected on routine urine toxicology screens. Since Maine voters approved of a ballot question on November 8, 2016, the recreational adult use, retail sale and taxation of marijuana has been legal in Maine ("An Act to Legalize Marijuana", Title 28-B, 128th legislature, 2016). This recent legislative change may account for some of the increase in the diagnosis of cannabis use at delivery due to either a true increase in use leading up to the new law or an increased comfort in disclosing use over time. Our finding of 64% of deliveries occurring with a diagnosis of nicotine use in this predominantly rural state is near the high end of the range of national prevalence estimates of tobacco use at the time of delivery among women with OUD, which varied from 2% overall[7] to 62% among rural women who delivered in a rural hospital.[1] Our estimate generally concurs with a study among pregnant women receiving medication-assisted treatment in Maine, which found a prevalence of tobacco use between 73 and 82%.[33]'

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* Corresponding author. University of Southern Maine, Muskie School of Public Service, 414 Wishcamper Center, 34 Bedford Street, Portland, ME, 04101, USA. Tel.: +207 780 4847; fax: +207 228 8138.
E-mail address: Katherine.ahrens@maine.edu (K.A. Ahrens).

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Table 1
Characteristics of delivery hospitalizations in Maine, 2009–2018 (n = 120,764).

	2009–2011		2012–2014		2015–2018	
	n = 37,503	%	n = 36,143	%	n = 47,118	%
Maternal Age (years)						
<20	2819	7.5	2114	5.9	2078	4.4
20–24	9524	25.4	8387	23.2	9415	20.0
25–29	11,385	30.9	11,158	30.9	14,659	31.1
30–34	8848	23.6	9514	26.3	13,674	29.0
>35	4927	13.1	4970	13.8	7292	15.5
State of Maternal Residence						
Maine	37,011	98.7	35,548	98.4	46,545	98.8
Other state/international/military	471	1.3	582	1.6	564	1.2
Missing	21	0.1	13	0.0	9	0.0
Rural-Urban Maternal Residence[a]						
All metro	13,184	35.2	12,690	35.1	16,666	35.4
Large rural	13,360	35.6	12,842	35.5	17,041	36.2
Small rural	8708	23.2	8226	22.8	10,460	22.2
Isolated	1758	4.7	1786	4.9	2368	5.0
Other state/international/military	493	1.3	597	1.7	581	1.2
Missing	0	0.0	2	0.0	2	0.0
Delivery Hospital Level of Care and Type						
Hospitals with level III NICU	12370	33.0	12,668	35.1	17,788	37.8
Hospitals with level II specialty care	4404	11.7	2911	8.1	2588	5.5
Hospitals with level I care, not critical access	16,040	42.8	16,523	45.7	22,285	47.3
Hospitals with level I care, critical access	5689	12.5	4041	11.2	4457	9.5
Delivery Type and Outcome						
Cesarean section	11,462	30.6	11,048	30.6	13,235	28.1
Stillborn	172	0.5	182	0.5	249	0.5
Contraception at Delivery Hospitalization						
Intrauterine device	115	0.3	143	0.4	48	0.1
Implant	[d]	[d]	[d]	[d]	162	0.3
Sterilization	2482	6.6	2391	6.6	2651	5.6
Maternal Co-occurring Conditions[b]						
Anxiety	809	2.2	1666	4.6	3773	8.0
Major depression	1370	3.7	1867	5.2	2831	6.0
Alcohol abuse or dependence	28	0.1	40	0.1	83	0.2
Other drug abuse or dependence[c]	81	0.2	52	0.1	212	0.5
Cannabis use	462	1.2	799	2.2	1706	3.6
Nicotine use	4758	12.7	4620	12.8	5470	11.6
Hepatitis C	193	0.5	306	0.9	508	1.1

Abbreviation: NICU, Neonatal intensive care unit.
[a] Rural-urban designations were determined using a four-category classification based on 2010 rural-urban community area codes (RUCAs), a census tract–based classification system: all metro (1, 1.1); large rural (2, 2.1, 3, 4, 4.1, 5, 5.1, 6); small rural (7, 7.1, 7.2, 8, 8.1, 8.2, 9, 10.1, 10.2, 10.3); and isolated rural (10).
[b] Maternal conditions and behaviors are not mutually exclusive; more than one may occur in an individual.
[c] Non-opioid and non-alcohol abuse or dependence.
[d] Cell size <10, values suppressed in accordance with data use agreement with Maine Health Data Organization.

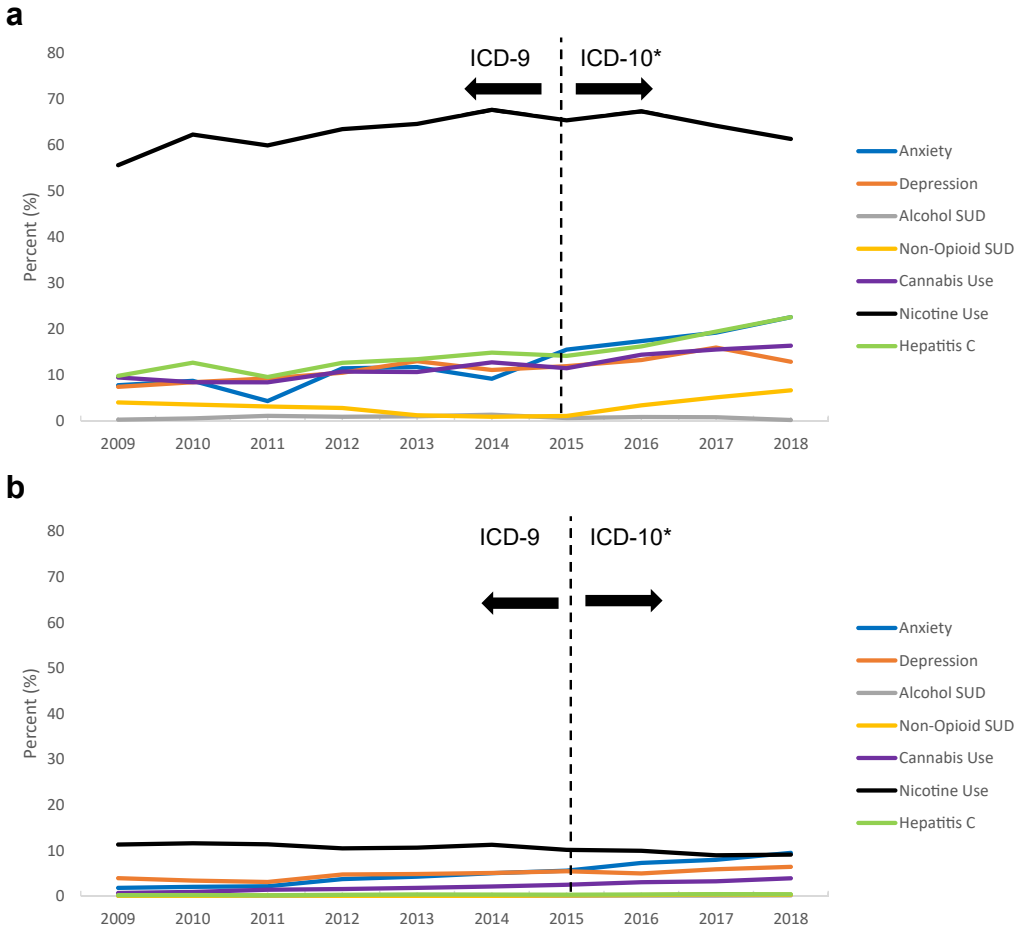
Table 3
Selected maternal co-occurring conditions among delivery hospitalizations in Maine with and without opioid use disorder, 2009–2018 (n = 120,764).

	Deliveries among Women with Opioid Use Disorder		Deliveries among Women without Opioid Use Disorder		PR	95% CI	P[a]
	n = 4026	% = 3.3	n = 116,738	% = 96.7			
Anxiety	534	13.3	5714	4.9	2.7	(2.5, 2.9)	<0.0001
Major depression	471	11.7	5597	4.8	2.4	(2.2, 2.7)	<0.0001
Alcohol abuse or dependence	34	0.8	117	0.1	8.4	(5.8, 12.3)	<0.0001
Other drug abuse or dependence[b]	128	3.2	217	0.2	17.1	(13.8, 21.2)	<0.0001
Cannabis use	486	12.1	2481	2.1	5.7	(5.2, 6.2)	<0.0001
Nicotine use	2557	63.5	12291	10.5	6.0	(5.9, 6.2)	<0.0001
Hepatitis C	598	14.9	409	0.4	43.4	(37.5, 47.9)	<0.0001

Abbreviation: CI, Confidence interval; PR, Prevalence ratio.
[a] Chi-square test P-value comparing deliveries of women with opioid use disorder to deliveries of women without opioid use disorder.
[b] Non-opioid and non-alcohol abuse or dependence.

SUBMITTING CORRECTED VERSIONS OF THE FOLLOWING:

Fig. 2 a Percent of co-occurring conditions at delivery hospitalizations among women with opioid use disorder (n = 4026). Fig. 2b Percent of co-occurring conditions at delivery hospitalizations among women without opioid use disorder (n = 116,738). *Vertical dashed line indicates data transition from ICD-9 to ICD-10, with 2015 being the first data year affected by the transition (which occurred on October 1, 2015).



*Vertical dashed line indicates data transition from ICD-9 to ICD-10, with 2015 being the first data year affected by the transition (which occurred on October 1, 2015).

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Original Research

COVID-19 fatality in Mexico's indigenous populations

A.D. Argoty-Pantoja [a], K. Robles-Rivera [a], B. Rivera-Paredez[*], J. Salmerón

Research Center in Policy, Population and Health, School of Medicine. National Autonomous University of Mexico, Mexico City, Mexico

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ABSTRACT

Objective: The aim of the study was to explore the factors that could explain the differences in fatality rates among indigenous groups with COVID-19 diagnosis compared with the rest of the population in Mexico.
Study design: We analyzed the public data of COVID-19 surveillance, of the Mexican Ministry of Health, to estimate COVID-19 fatality rates by ethnicity.
Methods: We explored associated factors using Cox proportional hazards models stratified by outpatient and hospital management at diagnosis; analysis was conducted in three scenarios: national level, states with 89% of the indigenous population, and South Pacific region.
Results: A total of 412,017 COVID-19 cases were included, with 1.1% of the indigenous population. The crude fatality rate per 1000 person-weeks was 64.8% higher among indigenous than among non-indigenous people (29.97 vs. 18.18, respectively), and it increased more than twice within outpatients (5.99 vs. 2.64, respectively). Cox analysis revealed that indigenous people who received outpatient management had higher fatality rate than non-indigenous outpatients, at the national level (hazard ratio [HR] = 1.63; 95% confidence interval [CI] = 1.34–1.98), within the subgroup of 13 states (HR = 1.66; 95% CI = 1.33–2.07), and in the South Pacific region (HR = 2.35; 95% CI = 1.49–3.69). Factors associated with higher fatality rates among non-indigenous and indigenous outpatients were age, sex, and comorbidities.
Conclusions: COVID-19 fatality is higher among indigenous populations, particularly within cases managed as outpatients.

Introduction

The severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) pandemic has more serious repercussions in vulnerable groups: older people with comorbidities, homeless people, pregnant women, and ethnic minority groups.[1–3] There are more than 476 million indigenous people in the world, which represent around 6% of the worldwide population,[4,5] and in Mexico, it is 10% of the total population.[6,7]

Indigenous populations are frequently affected by various crises owing to the economic and social conditions they live in. Their communities are usually isolated or poorly communicated, with poor access to health services. In many cases, such health services have little capacity and limited coverage, which may delay seeking medical attention, complicating early management and, therefore, leading to greater risks of complications and mortality. Health disparities have been documented among ethnic minority groups that have a higher prevalence of metabolic disorders, such as diabetes.[8]

The living conditions of indigenous populations in Mexico could place them in a higher impact of the SARS-CoV-2 epidemic. The number of deaths can be used as a key indicator of the trajectory of COVID-19 in our country.[9–11] Various studies have identified factors associated with lower survival in patients with COVID-19: men, more than 65 years old, and the presence of chronic comorbidities.[12–14] Among indigenous populations, the COVID-19 fatality of 18.8% was reported, compared with 11.8% in the general population. Nevertheless, the causes and risk factors that may be associated with mortality were not analyzed.[15] It is necessary to investigate in more detail how the epidemic is differentially affecting indigenous populations owing to socio-demographic differences, comorbidities, and the type of management received.

* Corresponding author. Cto. Centro Cultural S/N, Edificio Centro de Investigación en Poilíticas, Población y Salud 2do Piso, Ciudad Universitaria, Coyoacán, Ciudad de México, 04510, Mexico. Tel.: +52 55 56 22 66 66x82355.
E-mail address: bereriveraparedez7@gmail.com (B. Rivera-Paredez).
[a] Both authors contributed equally to this manuscript.

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We aim to explore those factors that could explain the fatality differences between indigenous people with COVID-19 diagnosis compared with the non-indigenous Mexican population.

Methods

Study design, setting, and participants

We performed a longitudinal analysis using the public data of the COVID-19 information derived from the Epidemiological Surveillance System of Viral Respiratory Diseases of suspected cases identified by the healthcare system in Mexico.[16] The study population included those cases with a positive diagnosis for SARS-CoV-2 infection certified by the Institute of Epidemiological Diagnosis and Reference (InDRE), from February 27, 2020, when the first case in the country was officially reported, until July 30, 2020 (n = 424,637).

Definitions of suspected and confirmed COVID-19 cases

A suspected case was defined as a 'person of any age who had at least two of the following signs and symptoms in the last 7 days: cough, fever, or headache accompanied by either dyspnea, arthralgias, myalgias, sore throat, rhinorrhea, conjunctivitis, or chest pain,' and the confirmed case was the suspected case with a diagnosis confirmed by the InDRE.[17]

Outcome of interest

Fatality rate was defined as the ratio of the number of deaths, occurred within the cohort study of confirmed COVID-19 cases, and the person-time at risk.

Covariates

Variables of interest were age, sex, state of residence, presence of chronic obstructive pulmonary disease (COPD), asthma, immunosuppression, cardiovascular disease, chronic kidney disease, smoking, metabolic comorbidities (joint effect of diabetes, hypertension, and obesity), date of admission in the cohort study, number of days from the symptom onset to seeking care, and severity of the patient's condition at the time of seeking care. This variable was defined based on the type of management at diagnosis: (a) outpatient management (OM), (b) hospital management (HM), and (c) management in the intensive care unit (ICU) and/or with intubation and assisted ventilation.

Indigenous population was defined as all individuals who declared to speak an indigenous language.

Statistical analysis

We conducted a descriptive analysis of indigenous and non-indigenous populations based on their survival condition. The person-time of the fatality rate was expressed in person-weeks based on the date from the symptom onset until death. Statistical differences between non-survivor indigenous people vs. non-survivor non-indigenous people were tested using the immediate two-sample proportion test for categorical variables and the non-parametric Mann-Whitney U test for numerical variables.

To investigate risk factors of COVID-19 fatality, the hazard ratio (HR) and 95% confidence interval (CI) were calculated using the multivariable Cox proportional hazards regression models stratified by management at diagnosis. For variables that did not meet the proportional risk assumption, an interaction with time was performed.[18,19] From this multivariable model, we explored the statistical significance of three-way interaction terms (indigenous × sex × time, indigenous × age-groups × time, and indigenous × comorbid conditions × time).

To improve comparability among the population groups, associations of interest were evaluated in three scenarios: considering the entire national population in Mexico, considering the population within the 13 states that concentrate 89% of the indigenous population in Mexico as per the National Population Council (Oaxaca, Chiapas, Veracruz, Estado de México, Puebla, Yucatán, Guerrero, Hidalgo, Quintana Roo, San Luis Potosí, Ciudad de México, Michoacán, and Campeche), and considering only three states of the South Pacific where the largest proportion of indigenous people is concentrated (34% in Oaxaca, Chiapas, and Guerrero). We excluded 12,610 cases without indigenous language information. There were no statistically significant differences in age (60.6 vs. 61.7, respectively), sex (men 66.1% vs. 64.9%), and comorbidities conditions such as diabetes (37.7% and 38%, respectively), hypertension (42.5% vs. 43.8%), or COPD (4.6% vs. 4.8%) between excluded and included individuals. All analyses were performed using Stata 14.1 and GraphPad Prism 8.2. All *P*-values were two tailed, and a *P*-value <0.05 was considered statistically significant.

Results

Characteristics of test-positive cases for COVID-19

The average age of non-survivors in the non-indigenous population with COVID-19 was 61.7 years (standard deviation [SD] = 14.2), more than half were in the 35- to 64-year age range, compared with 63.3 years of age (SD = 13.4), and almost half of them were 65 years or older, in the indigenous population. In both groups, the majority were men. Most comorbidities were more frequent in non-survivors in both the non-indigenous and indigenous population: hypertension (43.9% vs. 39.1%, respectively), diabetes (38.1% vs. 36.5%), obesity (24.7% vs. 25.6%, respectively), COPD (4.8% vs. 7.6%), immunosuppression (2.7% vs. 2.6%), cardiovascular disease (5.3% vs. 4.6%), chronic kidney disease (6.9% vs. 5.5%), and smoking (8.3% vs. 7.1%), except for obesity (24.7% vs. 25.6%, respectively) and asthma (with higher prevalence in indigenous non-survivors) (Table 1). Considering all comorbidities, 64.4% of the indigenous people who died had one metabolic comorbidity at least, compared with 66.6% of the non-indigenous people who died; the most prevalent ones in both groups were diabetes + hypertension, hypertension, and diabetes (Table 1). Regarding initial medical management, the majority of survivors received OM (80.5% of non-indigenous vs. 69.5% of indigenous people). A lower percentage of non-indigenous patients required hospitalization than indigenous (17.9% and 27.7%, respectively), as well as management in the ICU and/or with intubation (1.6% vs. 2.8%, respectively).

Among non-survivors, the majority were hospitalized (69.2% of non-indigenous vs. 63.7% of indigenous people), followed by management in the ICU and/or with intubation (19.6% vs. 23%, respectively), and a lower percentage received OM (11.2% vs. 13.3%, respectively).

The time from the symptom onset to seeking medical attention, as well as death, was similar in indigenous and non-indigenous people. Finally, non-survivor indigenous people had an average time of 6.5 days (SD = 7.2) from the beginning of hospitalization to death, compared with 7.7 days (SD = 7.5) in non-indigenous people (Table 1).

Table 1
Characteristics of test-positive cases for COVID-19 and fatality in Mexico.

Characteristics	Non-indigenous population			Indigenous population			*P*-value
	Total	Survivors	Non-survivors	Total	Survivors	Non-survivors	
	407,548 (98.9)	362,562 (89.0)	44,986 (11.0)	4469 (1.1)	3701 (82.8)	768 (17.2)	
Age (years), mean (SD)	45.2 (16.4)	43.1 (15.5)	61.7 (14.2)	50.4 (17.4)	47.7 (16.9)	63.3 (13.4)	
<35	117,173 (28.8)	31.9	3.4	905 (20.2)	23.9	2.7	0.288
35–64*	236,051 (57.9)	58.6	52.7	2537 (56.8)	58.7	47.4	0.004
≥65*	54,324 (13.3)	9.5	43.9	1027 (23.0)	17.4	49.9	0.009
Sex, N (%)							
Women	191,078 (46.9)	48.3	35.1	1813 (40.6)	42.0	33.5	0.357
Men	216,470 (53.1)	51.7	64.9	2656 (59.4)	58.0	66.5	0.357
Diabetes, N (%)	65,047 (16.0)	13.3	38.1	974 (21.9)	18.9	36.5	0.365
COPD,* N (%)	6354 (1.6)	1.2	4.8	161 (3.6)	2.8	7.6	0.000
Asthma,* N (%)	10,926 (2.7)	2.8	2.0	125 (2.8)	2.6	3.8	0.001
Immunosuppression, N (%)	4897 (1.2)	1.0	2.7	58 (1.3)	1.0	2.6	0.865
Hypertension,* N (%)	80,723 (19.9)	16.9	43.9	976 (21.9)	18.4	39.1	0.008
Cardiovascular disease, N (%)	8676 (2.1)	1.7	5.3	100 (2.3)	1.8	4.6	0.390
Chronic kidney disease, N (%)	8165 (2.0)	1.4	6.9	97 (2.2)	1.5	5.5	0.128
Obesity, N (%)	76,674 (18.9)	18.1	24.7	892 (20.0)	18.9	25.6	0.566
Smoking, N (%)	29,590 (7.3)	7.2	8.3	274 (6.2)	6.0	7.1	0.232
Metabolic comorbidities,[a] N (%)							
None	250,667 (61.7)	65.2	33.4	2438 (54.8)	58.8	35.6	0.200
Hypertension*	32,045 (7.9)	7.1	14.3	369 (8.3)	7.5	12.0	0.071
Obesity	44,645 (11.0)	11.3	8.6	529 (11.9)	12.1	11.0	0.019
Diabetes	23,054 (5.7)	5.0	11.0	416 (9.4)	8.9	11.3	0.792
Obesity + hypertension	13,674 (3.4)	3.1	5.7	140 (3.2)	2.8	5.0	0.406
Diabetes + hypertension	23.669 (5.8)	4.5	16.6	335 (7.5)	5.9	15.6	0.460
Diabetes + obesity	7007 (1.7)	1.6	3.2	91 (2.1)	1.8	3.0	0.755
Diabetes + obesity + hypertension	11,216 (2.8)	2.2	7.2	130 (2.9)	2.2	6.5	0.341
Initial management, N (%)							
Outpatients*	296,675 (72.8)	80.5	11.2	2674 (59.9)	69.5	13.3	0.068
Hospitalization *	96,041 (23.6)	17.9	69.2	1513 (33.9)	27.7	63.7	0.001
Hospitalization and/or ICU and/or intubation*	14,728 (3.6)	1.6	19.6	279 (6.3)	2.8	23.0	0.019
[b]Time from symptom onset to seeking care (days)*	4.3 (3.3)	4.3 (3.3)	4.4 (3.5)	4.3 (3.2)	4.2 (3.0)	4.7 (3.8)	0.012
[b]Time from symptom onset to death (days)*	12.1 (8.0)	–	12.1 (8.0)	11.2 (7.2)	–	11.2 (7.2)	0.002
[b]Time from seeking care to death (days)*	7.7 (7.5)	–	7.7 (7.5)	6.5 (7.2)	–	6.5 (7.2)	<0.001

SD = standard deviation; ICU = intensive care unit.
**P*-value <0.05 when comparing between non-survivors in the indigenous and non-indigenous population. For categorical variables, the immediate two-sample proportion test was used, and for continuous variables, we used the Mann-Whitney U test.
[a] None = without obesity, diabetes, hypertension. Obesity, diabetes, and hypertension categories do not exclude other types of comorbidities.
[b] Mean (SD).

COVID-19 crude fatality

The COVID-19 crude fatality rate per 1000 person-weeks was 64.8% higher in the indigenous population than in the non-indigenous population. In the indigenous population, 768 deaths were identified in 25,621 person-weeks (crude fatality: 29.97; 95% CI = 27.82–32.17), whereas in the non-indigenous population, 44,986 deaths were identified in 2,474,472 person-weeks (crude fatality: 18.18; 95% CI = 18.01–18.34).

When stratifying the analysis by type of management at diagnosis, we observed that the indigenous population had a higher crude fatality rate in both outpatients and hospitalized patients, than among non-indigenous people. Furthermore, we observed a significant difference in outpatients, wherein the indigenous population had a crude fatality rate more than twice the rate among non-indigenous patients (6.0 vs. 2.6, respectively). These results were similar in the subgroup of the 13 states containing 89% of the total indigenous population (2.4 vs. 6.1, respectively) and in the South Pacific region (2.6 vs. 7.6, respectively). In addition, we observed differences in time from the symptom onset to seeking care (days) among non-indigenous outpatients and indigenous outpatients for the different regions, and at the national level and in the 13 states, we observed an average time of 4.2 days in the non-indigenous population and 3.9 in the indigenous population ($P < 0.01$); however, in the South Pacific region, we observed that indigenous people have a longer time seeking care than non-indigenous people (4.5 vs. 4.2, $P < 0.001$, respectively). Within the outpatient group, the men were the most affected ones, wherein indigenous people had a crude fatality rate of 132% more than non-indigenous people; when assessing age, indigenous people in the 35- to 64-year age range had a crude fatality rate 119% higher than non-indigenous people of the same age-group (Table 2).

COVID-19 fatality risk

The results from the Cox proportional hazards analysis showed that sex, age, and the presence of comorbidities (COPD, hypertension, obesity, diabetes, and chronic kidney disease) are associated with a higher COVID-19 fatality rate, both in outpatients and in hospitalized patients.

Ethnicity was associated with a higher COVID-19 fatality rate in individuals who received OM, but not in individuals who received HM, regardless of age, sex, and comorbidities. In outpatients, we found that being indigenous increases the COVID-19 fatality rate by 63% compared with being non-indigenous (HR = 1.63; 95% CI = 1.34–1.98). We also observed that age ≥65 years had the highest risk when compared with age less than <35 years (HR = 30.68; 95% CI = 26.41–35.63), and the risk fatality in men increases by 97% compared with women (HR = 1.97; 95% CI = 1.86–2.09).

When evaluating metabolic comorbidities, we found that the risk was higher in people with diabetes (HR = 3.15; 95% CI = 2.63–3.77). The risk increases in people with diabetes and

Table 2
COVID-19 crude fatality rate in initial outpatient and hospitalized managements.

Study population characteristics	National level				States with 89% of the indigenous population				Oaxaca, Chiapas, Guerrero			
	Non-indigenous population		Indigenous population		Non-indigenous population		Indigenous population		Non-indigenous population		Indigenous population	
	Outpatients	Hospitalized	Outpatients	Hospitalized	Outpatients	Hospitalized	Outpatients	Hospitalized	Outpatients	Hospitalized	Outpatients	Hospitalized
Total, n	296,675	110,873	2674	1795	163,485	67,166	2183	1533	18,921	6866	414	367
Deaths	5008	39,978	102	666	2691	24,498	82	576	320	2795	21	134
Person-week	1,896,871	577,600	17,019	8603	1,109,718	369,473	13,407	7138	121,741	33,101	2746	1964
Fatality rate (95% CI)[d]	2.6 (2.6–2.7)	69.2 (68.5–69.9)	6.0 (4.9–7.3)	77.4 (71.8–83.5)	2.4 (2.3–2.5)	66.3 (65.5–67.1)	6.1 (4.9–7.6)	80.7 (74.5–87.6)	2.6 (2.4–2.9)	84.4 (81.4–87.6)	7.6 (5.0–11.7)	68.2 (57.6–80.8)
[a]Time SSC (days)[e]	4.2 (3.3)	4.4 (3.5)	3.9 (2.9)	4.7 (3.5)	4.3 (3.4)	4.5 (3.5)	3.9 (2.9)	4.8 (3.5)	4.2 (2.6)	4.3 (3.1)	4.5 (2.8)	4.6 (3.1)
[b]Time SD (days)[e]	12.8 (8.7)	12.0 (7.9)	10.9 (7.7)	11.2 (7.1)	12.8 (8.9)	12.2 (8.01)	10.8 (6.6)	11.1 (7.0)	11.3 (7.2)	11.4 (7.6)	10.0 (6.5)	10.8 (6.7)
[c]Time SCD (days)[e]	7.7 (8.2)	7.7 (7.4)	6.3 (7.4)	6.5 (7.2)	7.6 (8.3)	7.8 (7.5)	6.3 (6.4)	6.5 (7.1)	6.1 (6.6)	7.0 (7.2)	5.0 (5.4)	6.1 (6.0)
Women, n	148,222	42,856	1159	654	81,266	25,075	927	554	9057	2482	181	104
Deaths	1702	14,104	25	232	879	8117	20	205	110	926	3	39
Person-week	937,202	224,884	7408	3112	546,022	140,028	5685	2483	57,353	12,014	1186	526
Fatality rate (95% CI)[d]	1.8 (1.7–1.9)	62.7 (61.7–63.8)	3.4 (2.3–5.0)	74.6 (65.6–84.8)	1.6 (1.5–1.7)	58.0 (56.7–59.2)	3.5 (2.3–5.5)	82.6 (72.0–94.7)	1.9 (1.6–2.3)	77.1 (72.3–82.2)	2.5 (0.8–7.8)	74.2 (54.2–101.6)
Men, n	148,453	68,017	1515	1141	82,219	42,091	1256	979	9864	4384	233	263
Deaths	3306	25,874	77	434	1812	16,381	62	371	210	1869	18	95
Person-week	959,669	352,717	9611	5491	563,696	229,445	7723	4655	64,388	21,087	1560	1438
Fatality rate (95% CI)[d]	3.4 (3.3–3.6)	73.4 (72.5–74.2)	8.0 (6.4–10.0)	79.0 (71.9–86.8)	3.2 (3.1–3.6)	71.4 (70.3–72.5)	8.0 (6.3–10.3)	79.7 (72.0–88.2)	3.3 (2.9–3.7)	88.6 (84.7–92.7)	11.5 (7.3–18.3)	66.0 (54.0–80.8)
Age <35 years, n	106,480	10,693	776	129	57,027	6664	620	93	6229	759	108	44
Deaths	205	1328	4	17	108	807	3	14	8	86	2	6
Person-week	672,857	72,612	4983	856	379,561	47,547	3891	592	39,774	4718	729	308
Fatality rate (95% CI)[d]	0.3 (0.3–0.3)	18.3 (17.3–19.3)	0.80 (0.30–2.13)	19.9 (12.3–31.9)	0.3 (0.2–0.3)	16.9 (15.8–18.2)	0.8 (0.2–2.4)	23.6 (14.0–39.9)	0.2 (0.1–0.4)	18.2 (14.8–22.5)	2.7 (0.7–11.0)	19.5 (8.7–43.4)
Age 35–64 years, n	170,255	65,796	1558	979	95,165	40,726	1267	845	11,159	3778	254	208
Deaths	2751	20,968	55	309	1522	13,221	41	268	162	1334	8	68
Person-week	1,108,272	369,875	10,124	5128	658,945	240,710	7919	4302	72,990	19,957	1695	1113
Fatality rate (95% CI)[d]	2.5 (2.4–2.6)	56.7 (55.9–57.5)	5.43 (4.17–7.07)	60.3 (53.9–67.4)	2.3 (2.2–2.4)	54.9 (54.0–55.9)	5.2 (3.8–7.0)	62.3 (55.3–70.2)	2.2 (1.9–2.6)	66.8 (63.3–70.5)	4.7 (2.4–9.4)	61.1 (48.2–77.5)
Age ≥65 years, n	19,940	34,384	340	687	11,293	19,776	296	595	1533	2329	52	115
Deaths	2052	17,682	43	340	1061	10,470	38	294	150	1375	11	60
Person-week	115,742	135,114	1912	2619	71,213	81,216	1598	2243	8977	8427	322	543
Fatality rate (95% CI)[d]	17.7 (17.0–18.5)	130.9 (128.9–132.8)	22.5 (16.7–30.3)	129.8 (116.7–144.4)	14.9 (14.0–15.8)	128.9 (126.5–131.4)	23.8 (17.3–32.7)	131.1 (116.9–146.9)	16.7 (14.2–19.6)	163.2 (154.7–172.0)	34.1 (18.9–61.6)	110.5 (85.8–142.3)

CI = confidence interval.
[a] Time SSC: time from the symptom onset to seeking care (days).
[b] Time SD: time from the symptom onset to death (days).
[c] Time SCD: time from seeking care to death (days).
[d] Crude fatality rate per 1,000 person-weeks.
[e] Mean (SD).

hypertension (HR = 3.58; 95% CI = 3.05–4.22), obesity (HR = 4.69; 95% CI = 3.53–6.23), and hypertension + obesity (HR = 5.57; 95% CI = 4.54–6.84) (Fig. 1) (Table 3). We found an interaction effect with time in most of comorbidities in outpatients; in all cases, the risk of mortality decreased eventually, for example, the risk in people with chronic kidney disease during the first week is 3.58, and every week, the risk decreased by 17%, that is, in the second week, the risk decreased to 2.97 (95% CI = 2.60–3.40), and in the third week, it was 2.47 (95% CI = 2.17–2.81).

Furthermore, we did not observe statistically significant differences among outpatients between non-indigenous and indigenous people in variables such as sex (HR = 1.96 vs. 2.18), age (35–64 years, HR = 6.32 vs. 5.8, and >65 years, HR = 30.26 vs. 18.75), obesity + hypertension (HR = 2.61 vs. 2.3), diabetes + obesity + hypertension (HR = 4.03 vs. 3.37), and time from the symptom onset to seeking care (HR = 1.04 vs. 1.03).

In contrast to outpatients, in hospitalized patients, the COVID-19 fatality rate in indigenous and non-indigenous populations was similar (HR = 1.01; 95% CI = 0.94–1.09). We observed a positive interaction with time and sex, age, and hypertension, higher being in the following age-groups: ≥65 years and 35–64 years, wherein the risk increased by 26% and 21%, respectively.

Excess fatality in the indigenous population that received OM was observed in the following three scenarios: HR = 1.63 at the national level (95% CI = 1.34–1.98), HR = 1.66 in the subgroup of the 13 states containing 89% of the total indigenous population (95% CI = 1.33–2.07), and HR = 2.35 in the South Pacific region (95% CI = 1.49–3.69) (Fig. 2). The three-way interactions for indigenous × demographic (sex, age-groups) × time and indigenous × comorbid conditions × time were not statistically significant (*P* value >0.05).

Discussion

Our data suggest that management of treatment is the main factor associated with the differences in the COVID-19 fatality rates between the indigenous and non-indigenous population in the three scenarios (at the national level, in the subgroup of 13 states with 89% of the indigenous population, and in the South Pacific region). We observed that the indigenous population had a 64.8% higher crude fatality rate than non-indigenous people. Similar findings have been recorded in various countries, where it has been observed that ethnic minorities have a higher risk of dying from COVID-19. In Brazil, for example, the Pardo indigenous group was the second most important risk factor (after age) for death.[20]

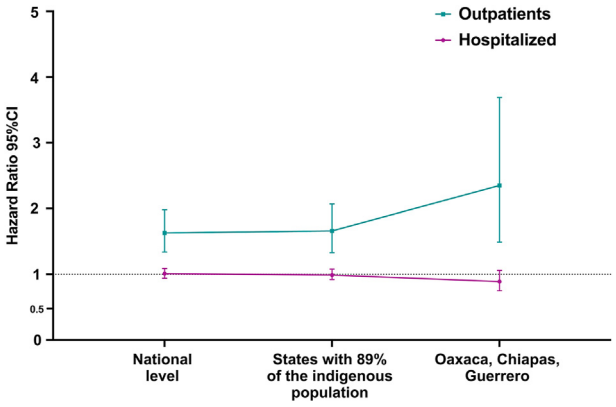

Fig. 1. COVID-19 fatality hazard ratios based on the type of management and the presence of comorbidities (multivariate model). HR reference: none (hypertension, obesity, diabetes). HR = hazard ratio; CI = confidence interval.

Similarly, the mortality rate in the United States of America is higher among Black people, Hispanics, or Asians, than in the white population.[21] In addition, in England and in Wales, ethnic disparities with regard to COVID-19 mortality have been observed: Black people, Indians, Pakistanis, Bangladeshis, and other ethnic groups had significantly higher risk of dying than the white population.[22]

In our data, after adjusting for sex, age, and metabolic comorbidities, the fatality rate is particularly higher among indigenous outpatients than among non-indigenous outpatients, whereas the fatality rates in hospitalized patients (indigenous and non-indigenous) are the same, in the three regions in Mexico (national, 13 states, and South Pacific region). Similar results were found in Georgia, USA, where the fatality rate during hospitalization was similar between African-Americans and other ethnic groups.[23]

When analyzing the differences in the prevalence of various comorbidities, it was found that non-survivor indigenous people had a higher frequency of comorbidities, being most affected by chronic and metabolic diseases, corresponding to the elevated prevalence of metabolic syndrome, central obesity, and hypertension in indigenous communities in Mexico.[24]

Historically, the indigenous population has shown poor health indicators in high rates of morbidity, disability, and early mortality, which are related to their own social, environmental, geographic, and cultural conditions. Access barriers are well-known factors that affect health results of these communities.[9,10,25] Unfortunately, the data set we used for this analysis is only a public administrative information, we acknowledge the data set lacks variables that measure access to care precisely, so we used time from the beginning of symptoms and seeking medical attention as the proxy variable. Nonetheless, in our study, we did not observe differences between non-indigenous and indigenous populations (4.3 vs. 4.3 h) regarding the chance to access medical attention. Furthermore, non-relevant differences were observed between time from the symptom onset and death in non-survivor indigenous and non-indigenous people (4.7 vs. 4.4, approximately 7 h).

Previous studies in different populations have documented that the person's perception of risk is important and is associated with the uptake of preventive and/or avoidant behaviors, which reported moderate risk perceptions in American, Australian, and UK individuals.[26–28] Among French individuals with high risk of severe COVID-19 (e.g., age >70 years and presence of chronic diseases), about 20% of them did not feel at risk and could therefore adopt avoidant behaviors.[29] We were unable to evaluate these factors in our analysis, but we consider this should be evaluated in further studies.

Despite the large volume of research on the pandemic, studies aimed at analyzing the association between ethnicity and COVID-19 are limited.[30] According to our knowledge, this is the first study in Mexico that analyzes COVID-19 fatality risk in the indigenous population. Although the number of national indigenous individuals screened for SARS-CoV-2 is small (n = 8835), it was possible to establish that they have higher COVID-19 fatality rates. These results, however, should be interpreted with caution as the nature of the data does not allow full understanding of the phenomenon that occurred in the indigenous population with COVID-19 and because of the observed underrepresentation as well.

Overall, our findings suggest that COVID-19 fatality is adversely affecting the indigenous population, particularly patients who received initial outpatient care. In addition, comorbidity mainly affects the indigenous population. Further analysis of the factors that could better explain the differential impact of COVID-19 in the indigenous population is warranted. In the meantime, an alternative may be to promote hospitalized management among indigenous populations. This may reduce disparities without increasing

Table 3
COVID-19 fatality hazard ratios with regard to initial outpatient and hospitalized management at the national level.

Study variables	Outpatients	Hospitalized
	HR (95% CI)	HR (95% CI)
Indigenous (reference: no)		
Yes	1.63 (1.34–1.98)	1.01 (0.94–1.09)
Sex (reference: women)		
Men	1.97 (1.86–2.09)	1.13 (1.09–1.18)[a]
Age (reference: <35 years)		
35–64 years	6.41 (5.55–7.40)	1.86 (1.68–2.07)[a]
≥65 years	30.68 (26.41–35.63)	3.16 (2.84–3.52)[a]
COPD (reference: no)		
Yes	2.19 (1.73–2.77)[a]	1.26 (1.16–1.37)[a]
Metabolic comorbidities (reference: none)		
Hypertension	2.20 (1.88–2.59)[a]	1.13 (1.06–1.20)[a]
Obesity	2.10 (1.74–2.53)[a]	1.13 (1.05–1.21)
Diabetes	3.15 (2.63–3.77)[a]	1.33 (1.25–1.41)
Obesity + hypertension	2.84 (2.29–3.51)	1.31 (1.21–1.42)
Diabetes + hypertension	3.58 (3.05–4.22)[a]	1.51 (1.43–1.59)[a]
Diabetes + obesity	4.69 (3.53–6.23)[a]	1.32 (1.18–1.46)
Diabetes + obesity + hypertension	5.57 (4.54–6.84)[a]	1.66 (1.54–1.79)[a]
Chronic kidney disease (reference: no)		
Yes	3.58 (2.88–4.44)[a]	1.93 (1.79–2.08)[a]

COPD = chronic obstructive pulmonary disease; HR = hazard ratio; CI = confidence interval.
[a] Interaction with time.

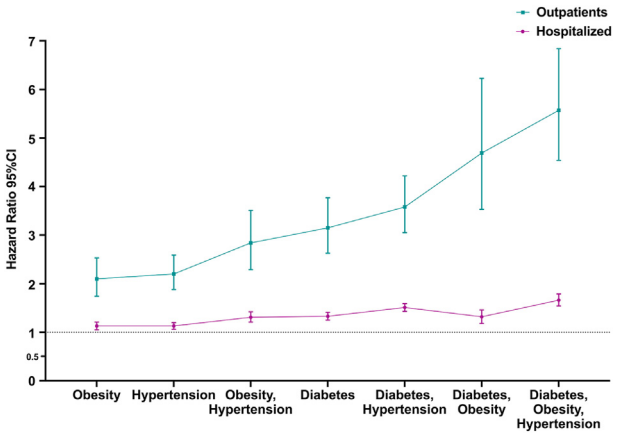

Fig. 2. COVID-19 fatality hazard ratios among indigenous people vs. non-indigenous people based on the type of management, in different regions in Mexico (multivariate model). HR reference: non-indigenous. States with 89% of the indigenous population: Campeche, Chiapas, Mexico City, Guerrero, Hidalgo, Estado de México, Michoacán, Oaxaca, Puebla, Quintana Roo, San Luis Potosí, Veracruz, and Yucatán. Three states in the South Pacific with the highest proportion of indigenous people: Oaxaca, Chiapas, and Guerrero. HR = hazard ratio; CI = confidence interval.

the healthcare service capacity overload, given the relatively small number of indigenous cases. Besides, health authorities mostly implement special care protocols for indigenous patients to reduce their fatality rates.

Author statements

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Ethical approval

No ethical approval was required as all the data analyzed were publicly available.

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None.

Competing interests

Nothing to disclose.

Author contributions

A.D.A.-P., K.R.-R., B.R.-P., and J.S. contributed to conception, design, data analysis, data interpretation, and manuscript writing. All authors reviewed the manuscript, had primary responsibility for final content, and read and approved the final manuscript.

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Original Research

Decomposition analysis of the compositional and contextual factors associated with poor-non-poor inequality in diarrhoea among under-five children in low- and middle-income countries

A.F. Fagbamigbe [a, b, *], O.P. Ologunwa [c], E.K. Afolabi [d], O.S. Fagbamigbe [e], A.O. Uthman [b]

[a] Department of Epidemiology and Medical Statistics, Faculty of Public Health, College of Medicine, University of Ibadan, Ibadan, Nigeria
[b] Division of Health Sciences, Populations, Evidence and Technologies Group, Warwick Medical School, University of Warwick, Coventry, United Kingdom
[c] Department of Economics, School of Management Technology, Federal University of Technology, Akure, Nigeria
[d] Department of Nursing Science, Faculty of Basic Medical Sciences, College of Health Sciences, Obafemi Awolowo University, Ile Ife, Nigeria
[e] Portsmouth Business School, Faculty of Business and Law, University of Portsmouth, Portsmouth, United Kingdom

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ABSTRACT

Objectives: The aim of the study was to assess the magnitude of wealth inequalities in the development of diarrhoea among under-five children in low- and middle-income countries (LMICs) and to identify and quantify contextual and compositional factors' contribution to the inequalities.
Design: This is a cross-sectional study.
Methods: We used cross-sectional data from 57 Demographic and Health Surveys conducted between 2010 and 2018 in LMICs. Descriptive statistics were used to understand the gap in having diarrhoea between the children from poor and non-poor households and across the selected covariates using Fairlie decomposition techniques with multivariable binary logistic regressions at $P = 0.05$.
Results: Of the 57 countries, we found a statistically significant pro-poor odds ratio in only 29 countries, 7 countries showed pro-non-poor inequality and others showed no statistically significant inequality. Among the countries with statistically significant pro-poor inequality, the risk difference was largest in Cameroon (94.61/1000), whereas the largest pro-non-poor risk difference in diarrhoea was widest in Timor-Leste (−41.80/1000). Important factors responsible for pro-poor inequality varied across countries. The largest contributors to the pro-poor inequalities in having diarrhoea are maternal education, access to media, neighbourhood socio-economic status, place of residence, birth order and maternal age.
Conclusion: Diarrhoea remains a major challenge in most LMICs, with a wide range of pro-poor inequalities. These disparities were explained by both compositional and contextual factors, which varied widely across the countries. Thus, multifaceted geographically specific economic alleviation intervention may prove to be a potent approach for addressing the poor and non-poor differentials in the risk of diarrhoea with policies tailored to country-specific risk factors. There is a need for further investigation of factors that drive pro-non-poor inequalities found in 9 of the LMICs.

Introduction

Diarrhoea is one of the major causes of morbidity and mortality among children, particularly in low- and middle-income countries (LMICs).[1] It is the second leading cause of deaths among under-five children (U5C), after acute respiratory infections, particularly pneumonia.[2–6] Globally, it is estimated that about 1.7 billion episodes of childhood diarrhoeal disease occur annually and more than 700,000 of these cases result in preventable deaths.[2,5–7] Thus, diarrhoea, especially among U5C, constitutes a public health concern and is a major obstacle to the achievement of the

Abbreviations: BODA, Blinder-Oaxaca decomposition analysis; CI, confidence interval; DHS, Demographic and Health Survey; FDA, Fairlie decomposition analysis; IRB, Institutional Review Board; LMIC, low- and middle-income countries; OR, odds ratio; PSU, primary sample unit; RD, risk difference; SES, socio-economic status; U5C, under-five children; UNICEF, United Nations International Children's Emergency Fund; WHO, World Health Organization.

* Corresponding author. Department of Epidemiology and Medical Statistics, Faculty of Public Health, College of Medicine, University of Ibadan, Ibadan, Nigeria Tel.: +234806134865.
E-mail address: franstel74@yahoo.com (A.F. Fagbamigbe).

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Sustainable Development Goal (SDG) on the reduction of child mortality in LMICs.

Although the contest against diarrhoea among U5C was the subject of several international, national and regional interventions in LMICs, diarrhoea occurrence has persisted in these countries and the poor-rich divide may have widened.[8] It has been estimated that every child has at least 4 episodes of diarrhoea per year in South-east Asia and Africa.[4,9] High prevalence of diarrhoea, as high as 35%, has been reported across these regions, reaching with both seasonal fluctuations and spatial variations.[10] These regions constitute more than 90% of the LMICs.

Diarrhoea is considered a symptom of wider socio-economic inequality within and across populations.[11] Diarrhoea has long-term negative effects on individuals' countries' socio-economic development.[1] Income inequality had been existent for a long time and is regarded as a strong indicator of health uptake.[12] The wider the gap between the rich and the poor in a given area, the worse the health outcomes of that area, a phenomenon that may result in less social cohesion and greater psychosocial stress on the child's growth and well-being that has detrimental health implications among the vulnerable population.[13] Increase in socio-economic inequality has potential impact on the distribution of household- and regional-level determinants of child health outcomes, including diarrhoea.[14] Socio-economic inequalities in health persist among children from poorer regions and neighbourhoods because they have a higher likelihood of exposure to conditions that exacerbate health outcomes.[15]

Earlier studies have documented factors that significantly contributed to having diarrhoea among U5C.[11,13,16–19] These factors include childhood and maternal deprivation, environmental sanitation, maternal health-seeking behaviour and ethnicity diversity.[20] Dairo et al.[4] reported improper disposal of faeces and contaminated water and food as major risk factors of diarrhoea. These factors gave credence to the fact that most populations of the world, especially in LMICs, are still afflicted by poverty, poor sanitation and lack of hygiene.[21–23]

However, these studies have been limited in scope to regions, within-country and inter-district analysis. Besides, we are not aware of any study that focused on identifying compositional and contextual factors that contribute to wealth inequalities in having diarrhoea among U5C in LMICs as a whole, whereas a good understanding of the magnitude and determinants of wealth inequalities in the development of diarrhoea may help reduce these inequalities and prevent diarrhoea occurrences and in reaching the SDGs on reduction in child morbidity and mortality.[24] It is therefore imperative to understand the effect of poor-non-poor inequalities on diarrhoea and identify the drivers of the inequality in LMICs. Besides, the identification of these factors could help inform the focus, levels, direction and magnitude of interventions targeted at closing wealth-related gaps in the prevention of diarrhoea among U5C in LMICs.

The study objectives are to assess the magnitude of wealth inequalities in the development of diarrhoea among U5C in LMICs, identify the compositional and contextual factors that contribute to pro-poor inequalities and quantify the contributions of the significant factors to the inequalities. As postulated by Kumi-Kyereme and Amo-Adjei,[25] we hypothesised that children from poorer households will have a higher likelihood of developing childhood diarrhoea. The study outcomes will provide comparative and evidence-based information that will assist policymakers, program implementers and stakeholders in intervention strategies to address the effect of poor-non-poor inequalities in the development of diarrhoea in LMICs.

Methods

Study design and data

We used data from the Demographic and Health Surveys (DHSs) collected periodically across the LMICs.[26–30] The DHS is cross-sectional nationally representative population-based household surveys. We pooled data from the most recent successive DHS conducted within the last ten years (2010–2019) and available as of April 2020 when our data curation took place. In addition, the survey must have captured information on diarrhoea experience among U5C. Only 57 LMICs met this inclusion criterion and were thus analysed in this study. In each of the countries, the DHS used a multistage stratified sampling design (usually from states/divisions/regions to district to clusters). The households were then selected from the clusters that are the primary sampling units (PSUs).[31,32]

Dependent variable

The outcome variable in this study is the recent experience of diarrhoea. Diarrhoea is defined as 'passage of liquid stools three or more times a day'[33,34] and 'recent experience of diarrhoea' as having an episode of diarrhoea within two weeks before the interview date.[28] The mothers were asked to list their U5C. For each child, the mother was asked if the child had diarrhoea within two weeks preceding the survey. The responses were binary: yes or no.

Main determinant variable

The main determinate variable in this decomposition study is poverty: poor or non-poor. Owing to non-availability of data on participants' expenditures and incomes, the DHS recommended and used household asset ownership as a proxy for calculating household wealth status, which can then be interpreted as an indicator of children households' poverty status. The household wealth quintiles are computed as a composite score of assets owned by households.[35] Additional details of the methodologies and country-specific assets used for the computation of the wealth quintiles are http://dhsprogram.com.[36] The DHS data have already generated and categorised the household wealth quintile variable into 5 categories of 20% each. In this study, we recategorised the household wealth quintile into two categories, poor (lower 40%) and non-poor (upper 60%), so that we can compare recent experiences of diarrhoea among U5C from poor and non-poor groups. A similar categorisation has been used elsewhere.[37–40] Hence, we defined 'wealth inequality' as 'the unequal distribution of assets' among households.

Independent variables

These are made up of the individual-level and neighbourhood-level factors as identified in the literature.[4,11,13,16–21,38–40]

Individual-level factors

The individual-level factors (compositional factors) consist of childs', mothers' and households' characteristics. Childs' characteristics were as follows: sex (male versus female), age in years (less than 1 year and 12–59 months), weight at birth (average+, small and very small), birth interval (firstborn, <36 months and >36 months) and birth order (1, 2, 3 and 4+). Mothers' characteristics were as follows: maternal education (none, primary or secondary plus), maternal age (15–24, 25–34, 35–49), marital status (never, currently and formerly married) and employment status (working

Table 1
Description of demographics and health surveyed data by countries, poverty and diarrhoea prevalence among under-five children in LMICs, 2010–2018.

Country	Year of survey	Number of clusters	Number of under-five children	Weighted (%) poor	Weighted diarrhoea prevalence (%)		
					Overall	Poor	Non-poor
All		63,378	796,150	44.6	14.2[b]	15.0[a]	13.6
Eastern Africa		6298	102,886	45.2	16.7	17.6[a]	15.9
Burundi	2016	554	12,431	43.4	22.5	24.7[a]	20.8
Comoros	2012	252	2949	45.7	17.0	17.8	16.3
Ethiopia	2016	643	9916	46.9	11.9	11.1[a]	12.6
Kenya	2014	1593	19,889	44.3	15.4	17.3[a]	13.8
Malawi	2016	850	16,246	47.1	21.9	22.8[a]	21.2
Mozambique	2011	610	10,157	45.0	11.2	11.1	11.4
Rwanda	2014	492	7474	45.6	12.2	14.7[a]	10.2
Tanzania	2015	608	9445	45.9	12.1	10.4[a]	13.6
Uganda	2016	696	14,379	43.6	20.0	22.0[a]	18.4
Middle Africa		3081	71,630	44.0	19.0	19.6[a]	18.5
Angola	2016	625	13,463	45.1	15.7	14.9[a]	16.4
Cameroon	2011	578	10,326	44.2	21.7	27.0[a]	17.5
Chad	2015	624	16,710	42.0	22.3	23.6[a]	21.3
Congo	2012	384	8723	46.3	19.3	17.5[a]	20.9
Congo DR	2014	536	16,994	43.9	17.0	16.5	17.4
Gabon	2012	334	5414	43.9	16.8	17.6	16.2
Northern Africa		874	15,458	37.5	14.0	16.4[a]	12.7
Egypt	2014	874	15,458	37.5	14.0	16.4[a]	12.7
Southern Africa		2544	25,529	44.9	15.5	17.1[a]	14.3
Lesotho	2014	396	2824	42.4	12.2	13.4	11.4
Namibia	2013	536	4449	43.9	19.1	22.7[a]	16.2
South Africa	2016	668	3241	45.0	11.0	13.8[a]	8.7
Zambia	2018	545	9311	47.5	15.5	16.4[a]	14.7
Zimbabwe	2015	399	5704	42.6	17.1	17.7	16.6
West Africa		6285	139,382	43.3	14.7	16.4[a]	13.4
Benin	2018	555	12,512	41.4	10.5	12.0[a]	9.5
Burkina Faso	2010	573	13,621	41.7	14.9	13.9[a]	15.5
Cote d'Ivoire	2012	351	6876	47.3	18.5	18.9	18.0
Gambia	2013	281	7633	42.4	17.8	17.0	18.3
Ghana	2014	427	5539	43.2	11.9	14.3[a]	10.0
Guinea	2015	401	7213	44.8	14.6	14.1	15.0
Liberia	2013	322	6806	46.5	22.7	24.9[a]	20.9
Mali	2018	345	9171	41.7	17.2	20.0[a]	15.3
Niger	2012	476	11,437	40.0	14.4	13.6	14.9
Nigeria	2018	1389	30,603	43.5	12.8	17.1[a]	9.6
Senegal	2017	400	11,253	46.2	18.0	20.6[a]	15.8
Sierra Leone	2013	435	10,254	45.5	11.5	11.3	11.7
Togo	2013	330	6464	41.3	15.2	19.4[a]	12.2
Central Asia		682	10,216	38.8	10.2	11.8[a]	9.1
Kyrgyz Republic	2012	316	4222	38.9	5.2	5.4	5.0
Tajikistan	2017	366	5994	38.8	13.3	15.9[a]	11.6
South-Eastern Asia		1850	17,168	47.5	9.0	10.0[a]	8.0
Cambodia	2014	609	6934	44.0	12.9	14.1[a]	11.8
Philippines	2017	1241	10,234	50.1	6.1	7.3[a]	4.9
Southern Asia		33,053	322,219	45.3	11.5	11.7[a]	11.4
Afghanistan	2015	956	30,520	39.7	29.1	27.6[a]	30.1
Bangladesh	2014	600	7541	41.4	5.7	6.3	5.3
India	2016	28,321	247,181	46.7	9.2	9.9[a]	8.6
Indonesia	2017	1967	17,155	40.5	14.2	16.1[a]	12.9
Maldives	2016	265	3048	41.9	4.2	4.3	4.2
Nepal	2016	383	4827	42.3	7.7	7.0	8.2
Pakistan	2018	561	11,947	42.0	19.2	18.3[a]	19.9
Western Asia		2048	27,441	46.1	21.8	22.5[a]	21.2
Armenia	2016	306	1709	39.4	3.8	4.9	3.1
Jordan	2017	962	10,454	50.8	9.7	10.1	9.2
Yemen	2013	780	15,278	43.9	31.4	33.3[a]	29.9
Central America		1996	22,524	47.0	18.7	19.5[a]	18.0
Guatemala	2014	856	12,038	48.7	19.2	18.9	19.6
Honduras	2011	1140	10,486	44.8	18.0	20.2[a]	16.2
South America		1401	9408	47.1	12.3	13.7[a]	11.1
Peru	2012	1401	9408	47.1	12.3	13.7[a]	11.1
Southern Europe		651	2745	44.0	6.1	7.7[a]	4.9
Albania	2018	651	2745	44.0	6.1	7.7[a]	4.9
Caribbean		1860	21,129	45.2	15.0	15.2	14.8
Dominican Republic	2013	516	3560	46.6	18.2	22.1[a]	14.7
Haiti	2016	449	6082	45.6	21.4	20.7	21.9
Myanmar	2014	440	4575	51.6	10.5	12.4[a]	8.4

(*continued on next page*)

Table 1 (*continued*)

Country	Year of survey	Number of clusters	Number of under-five children	Weighted (%) poor	Weighted diarrhoea prevalence (%)		
					Overall	Poor	Non-poor
Timor-Leste	2016	455	6912	40.4	10.8	8.3[a]	12.5
Oceania		755	8415	41.4	15.4	14.8[a]	15.9
Papua New Guinea	2016	755	8415	41.4	15.4	14.8[a]	15.9

LMIC = low- and middle-income country.
[a] Significant at 5% test of equality of proportions between poor and non-poor.
[b] Significant at 5% chi-squared test.

or not working). Households' characteristics were as follows: access to media (at least one of radio, television or newspaper), sources of drinking water (improved or unimproved), toilet type (improved or unimproved), cooking fuel (clean fuel or biomass) and housing materials (improved or unimproved). However, housing materials, access to toilet and clean water were excluded from the decomposition analysis because the DHS has already used them to compute the household wealth quintile, which is the main determinate variable in this study.

Neighbourhood-level factors

The DHS used 'clusters' as the PSU to group people of the same cluster that shares similar contextual factors.[31,32] We used the word 'neighbourhood' to describe the clustering of children within the same geographical environment and 'neighbours' as members of the same cluster. The PSUs were identified using the most recent census in each country where the DHS was held. In this study, we considered rural-urban residence and neighbourhood socioeconomic status (SES) as community-level variables. The neighbourhood SES was computed using principal component factor composed of the proportion of respondents within the same neighbourhood without education and employment.

Statistical analyses

Descriptive and inferential statistics comprising bivariable analysis and multivariable Fairlie decomposition techniques using binary logistic regressions were used. The z-test for equality of proportions of children who had diarrhoea from poor and non-poor households within each country and region was conducted and is reported in Table 1, whereas the existence of an association between the explanatory variables and the outcome variable among the two groups of children is reported in Table 2. The risk difference (RD) is the difference in the experience of diarrhoea among U5C from poor and non-poor households (Fig. 1). Charts were used to show the distributions of the RDs versus the prevalence of diarrhoea (Figs. 2 and 3). Finally, the adjusted logistic regression method was applied to the 29 pro-poor countries to carry out a Fairlie decomposition analysis, and the results are presented in Fig. 4.

Decomposition analysis

Multivariable decomposition is often used to quantify the contributions to differences in the prediction of an outcome of interest between two groups in multivariate models.[41] The Fairlie technique works by decomposing the difference in proportions based on either the probit or logit model.[41] The decomposition analysis is carried out by calculating the difference between the predicted probability for one group (say group A) using the other group's (say group B) regression coefficients and the predicted probability for that group (group A) using its regression coefficients.[42] The Fairlie decomposition technique works by constraining the predicted probability between 0 and 1.

Fairlie et al.[41] showed that the decomposition for a non-linear equation can be expressed as follows:

$$\overline{Y}^{A} - \overline{Y}^{B} = \overbrace{\left[\sum_{i=1}^{N^{A}} \frac{F\left(X_i^{A}\widehat{\beta}^{A}\right)}{N^{A}} - \sum_{i=1}^{N^{B}} \frac{F\left(X_i^{B}\widehat{\beta}^{A}\right)}{N^{B}}\right]}^{1^{st}} + \overbrace{\left[\sum_{i=1}^{N^{B}} \frac{F\left(X_i^{B}\widehat{\beta}^{A}\right)}{N^{B}} - \sum_{i=1}^{N^{B}} \frac{F\left(X_i^{B}\widehat{\beta}^{A}\right)}{N^{B}}\right]}^{2^{nd}} \quad (1)$$

where YJ is the average probability of the binary outcome variable with group J and F as the logistic cumulative distribution function, XJ is a row vector of the average values of the explanatory variables and βJ is a vector of coefficient estimates for group J. The numerical details have been reported.[43,44] NA is the sample size for group J.[45]

We used the 'Fairlie' Ado file in STATA to carry out the decomposition analysis using the generalised structure for the model. The R statistical software was used to draw all the figures. All the estimates were weighted, and all statistical tests were set to the 5% significance level. The results of this study are presented in Tables 1 and 2 and Figs. 1–4.

Results

The analysed data consist of 796,150 U5C living within 63,378 neighbourhoods nested in 57 LMICs. The overall proportion of children from poor households was 45%, the lowest in Egypt (38%) and highest in Myanmar (52%). The overall diarrhoea prevalence was 14.2% (significantly different across countries at $P < 0.001$), with 15.0% and 13.6% ($P < 0.001$) among children from poor and non-poor households, respectively (Table 1 and Fig. 1). The prevalence of diarrhoea among children from poor households ranged from 4.3% in Maldives to 33.3% in Yemen, whereas it ranged from 3.1% in Armenia to 30.1% in Afghanistan among children from non-poor households. The z-test of equality of prevalence among children from poor and non-poor households was statistically significant ($P < 0.05$) in 35 countries.

We found statistical significance in the association among all the explanatory variables considered in this study ($P < 0.05$) with the occurrence of diarrhoea and also by poverty divides of the children households, except media access and the sex of the household head that were insignificantly associated with the occurrence of diarrhoea among children from poor and non-poor households (Table 2). The prevalence of diarrhoea was consistently higher among the infants than among those aged 12–59 months, irrespective of their households' poverty status: 18% vs 14% for poor households and 17% vs 13% for non-poor households, respectively.

Table 2
Summary of pooled sample characteristics of the studied children in 57 LMICs.

Characteristics	N	Weighted %	Weighted (%) poor	Weighted diarrhoea prevalence (%)		
				Overall	Poor	Non-poor
Age						
Infant	164,438	20.7	44.4	17.4[a]	18.1[a]	16.9[a]
12–59 months	631,712	79.4	44.7	13.4	14.1	12.8
Sex						
Female	389,173	48.9	45	13.8[a]	14.6[a]	13.1[a]
Male	406,977	51.1	44.3	14.6	15.3	14.1
Household head						
Male	669,287	84.1	44.4	14.2[a]	14.9[a]	13.6
Female	126,863	15.9	45.8	14.5	15.5	13.7
Maternal age						
15–24 years	234,550	29.5	46.3	16.4[a]	16.6[a]	16.1[a]
25–34 years	414,014	52.0	42.5	13.2	14.1	12.6
35–49 years	147,586	18.5	47.9	13.4	14.4	12.5
Maternal education						
No education	273,056	34.3	62.7	15.8[a]	15.2[a]	16.6[a]
Primary	202,835	25.5	51.7	16.3	16.3	16.2
Secondary or higher	320,257	40.2	25.6	11.7	12.7	11.4
Employment						
Employed	526,983	66.2	45.6	13.3[a]	14.3[a]	12.4[a]
Unemployed	269,167	33.8	42.7	16.0	16.3	15.8
Media access						
No	316,993	39.9	67.1	15.2[a]	15.1	15.5[a]
Yes	478,517	60.2	30.8	14.2	14.8	13.1
Drinking water sources						
Unimproved sources	175,663	22.8	65.8	16.9[a]	17.2[a]	16.3[a]
Improved sources	595,332	77.2	39.2	13.6	14.0	13.3
Toilet type						
Unimproved sources	388,386	50.4	66.4	15.4[a]	15.2[a]	15.9[a]
Improved sources	382,305	49.6	23.6	13.1	14.5	12.7
Marital status						
Never married	23,560	3.0	37.7	16.9[a]	18.3[a]	16.0[a]
Currently married	739,740	92.9	44.7	14.0	14.7	13.4
Formerly married	32,850	4.1	47.3	17.1	17.9	16.4
Cooking fuel						
Unclean/biomass	581,710	77.0	56.2	14.9[a]	15.0	14.8[a]
Clean fuel	173,921	23.0	12.4	12.4	14.8	12.1
Housing materials						
Unimproved sources	676,227	89.5	49.5	14.8[a]	15.1[a]	14.6[a]
Improved source	79,157	10.5	12.2	10.0	10.7	9.9
Weight at birth						
Average+	643,472	84.0	43.5	13.6[a]	14.3[a]	13.1[a]
Small	90,809	11.9	47.9	17.2	18.3	16.2
Very small	31,924	4.2	50.6	20.1	20.7	19.4
Birth interval						
1st birth	223,779	28.2	37.3	13.1[a]	14.3[a]	12.4[a]
<36 months	308,310	38.8	51.0	15.0	15.3	14.7
36+ months	262,278	33.0	43.8	14.3	14.9	13.7
Birth order						
1st	223,777	28.1	37.3	13.1[a]	14.3[a]	12.4[a]
2nd	192,088	24.1	40.1	13.1	13.9	12.6
3rd	129,829	16.3	46.3	14.2	14.6	13.9
4+	250,456	31.5	54.4	16.2	16.2	16.1
Location						
Urban	239,222	30.1	14.5	13.4[a]	13.8[a]	13.3[a]
Rural	556,928	70.0	58.8	14.6	15.1	13.9
Neighbourhood SES						
Highest	159,709	20.1	18.7	9.8[a]	10.9[a]	9.6[a]
2	158,969	20.0	23.0	14.9	13.9	15.2
3	160,077	20.1	50.5	15.8	16.1	15.5
4	159,153	20.0	59.0	16.7	17.3	15.8
Lowest	158,242	19.9	74.9	14.0	13.6	15.1
Total	796,150	100.0	44.6	**14.2	**15.0	**13.6

LMIC = low- and middle-income country; SES = socio-economic status.
[a] (a) *significant at 5% test of equality of proportions between poor and non-poor (b) **a Significant at 5% chi-squared test.

Magnitude and variations in poor-non-poor inequality in diarrhoea

A meta-analysis of the RDs, a measure of inequality in the risk of having diarrhoea among children from poor and non-poor households, across the 57 countries is presented in Fig. 1. The prevalence of diarrhoea was generally higher among children from poor households than those from non-poor households in all the countries, except in Angola, Congo, Ethiopia, Burkina Faso, Pakistan, Afghanistan, Niger, Tanzania and Timor-Leste, where the RD was significantly higher among children from non-poor households. The differences were however insignificant in 19 countries. The overall, that is, random effect of the RD was 17.31 of 1000 children,

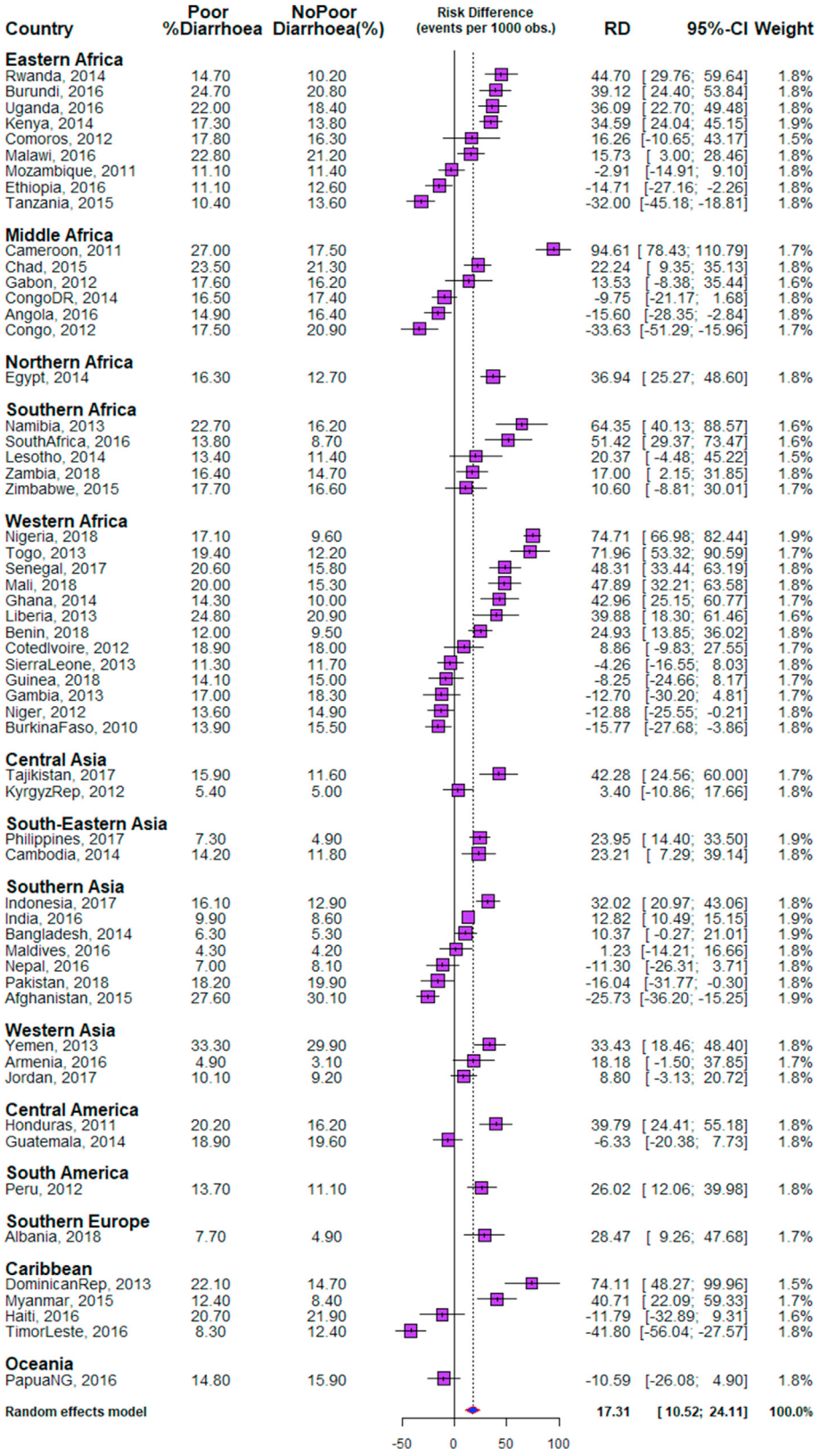

Fig. 1. Risk difference between children from poor and non-poor households in the prevalence of diarrhoea by countries. CI = confidence interval; RD = risk difference.

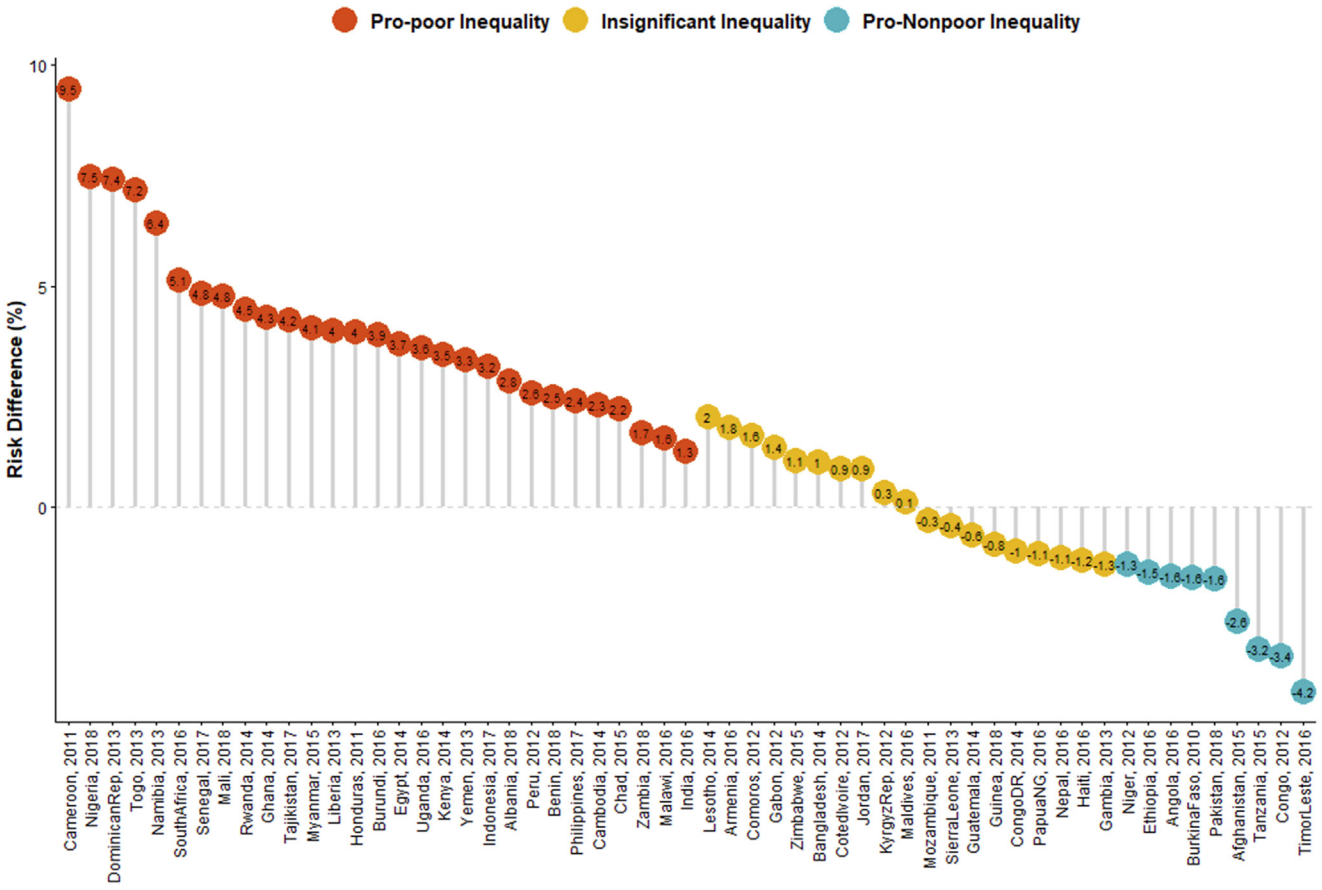

Fig. 2. Risk difference between children from poor and non-poor households in the prevalence of diarrhoea by countries.

with a 95% confidence interval (CI) of 10.52–24.11. This is evident of significant overall pro-poor inequality. The greatest contribution (weight) to the random effect was found in Kenya, Nigeria, Philippines, Bangladesh, Afghanistan and India, at 1.9% each, whereas the least contribution was found in Comoros, Lesotho and the Dominican Republic, at 1.5% each (Fig. 1).

Relationship between prevalence of diarrhoea and magnitude of inequality

The relationships between the prevalence of diarrhoea and the magnitude of poor-non-poor inequality, a function of RD, across the 57 countries are presented in Fig. 3. We categorised the countries into 4 distinct categories based on their prevalence of diarrhoea and whether or not the RD were small or large: (i) high diarrhoea prevalence and high pro-poor inequality countries such as Togo, Yemen, Cameroun, the Dominican Republic and Liberia; (ii) high diarrhoea prevalence and high pro-non-poor inequality countries such as Afghanistan, Congo and Pakistan; (iii) low diarrhoea prevalence and high pro-poor inequality countries such as Nigeria, South Africa, Rwanda and Tajikistan; and (iv) low diarrhoea prevalence and high pro-non-poor inequality countries such as Timor-Leste, Tanzania and Ethiopia.

Decomposition of poverty inequality in the prevalence of diarrhoea

We first computed Mantel-Haenszel pooled estimate of the odds ratio (OR) of having diarrhoea while controlling for the country among all the children as 1.12 (95% CI: 1.11–1.14) and tested the null hypothesis that OR = 1; we estimated z = 17.4 and $P = 0.000$ and Test of heterogeneity, we estimated $X^2 = 819.27$, degree of freedom (d.f.) = 56, and $P = 0.000$, I-squared (variation in OR attributable to heterogeneity) = 93.2%. Of the 57 countries, we found statistically significant pro-poor OR (pro-poor inequality) in only 29 countries, 7 showed pro-non-poor inequality and the remaining 21 countries showed no statistically significant inequality.

Across the 29 countries, the largest contributions to gaps in having diarrhoea among the groups of children from the poor and non-poor households are maternal education, access to media neighbourhood SES, place of residence, birth order and maternal age. Among these contributors, the maternal education and access to media contributed most and were clustered together, whereas the other important contributors formed another cluster (Fig. 4). Bangladesh, India, Malawi, Zambia and Peru had the highest experience of the contributions of these factors, as shown in the clustering in Fig. 4. The contributions were most visible in Bangladesh and India. Specifically, the largest contributions to pro-poor inequality in the prevalence of diarrhoea in Bangladesh were maternal education (306% higher among children whose parents had no education), media access (219% higher among children whose mother had no media access) and neighbourhood SES (170% higher in communities with lowest SES), followed by birth order (130%), maternal education (21%) and place of residence (19% higher among rural residents). Other factors such as childbirth weight, age, sex and mothers' employment status had the lowest contribution to poverty-related inequalities in the prevalence of diarrhoea across these countries.

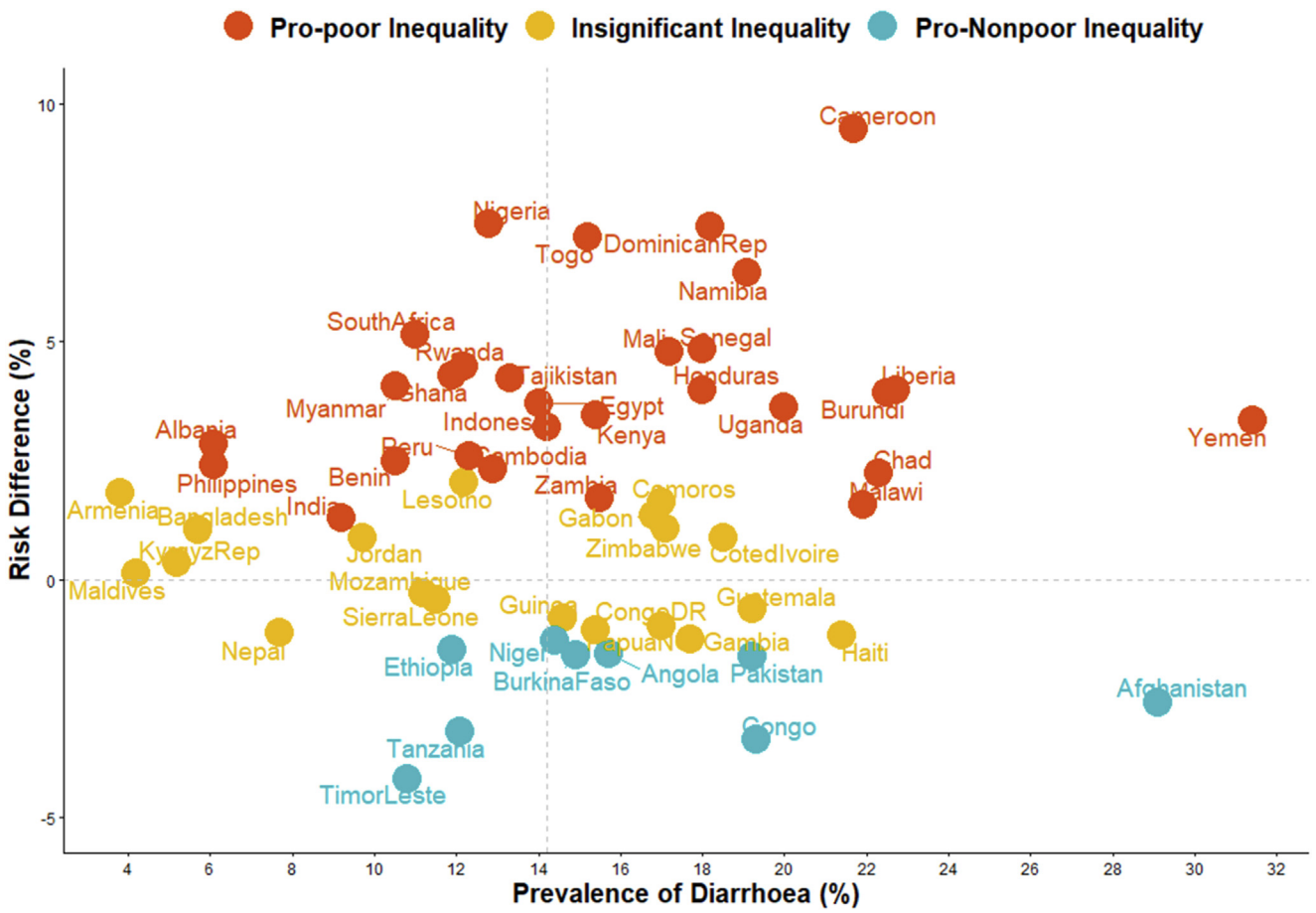

Fig. 3. Scatter plot of the rate of diarrhoea and risk difference between children from poor and non-poor households in LMICs. LMIC = low- and middle-income country.

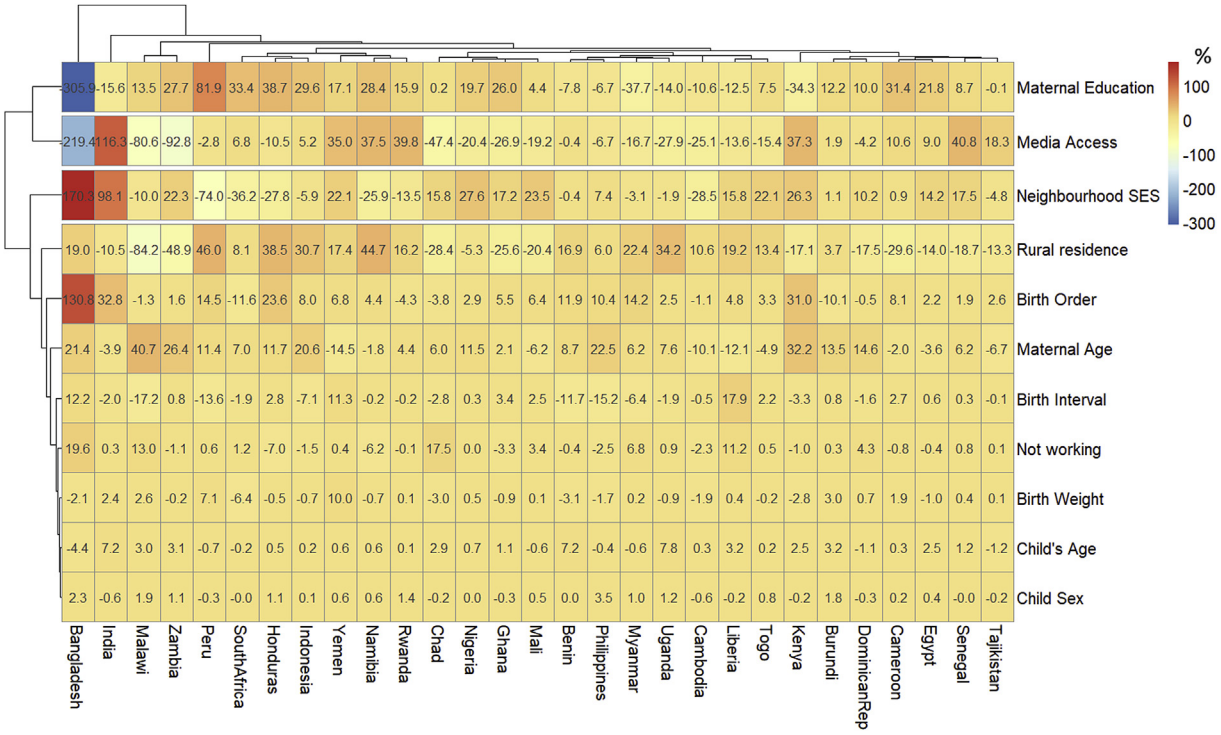

Fig. 4. Contributions of differences in the distribution 'compositional effect' of the determinants of diarrhoea to the total gap between children from poor and non-poor households by countries. SES = socio-economic status.

Discussion

The need to understand the compositional and contextual factors that contributed to the gap in the risk of diarrhoea among children from poor and non-poor households in 57 LMICs motivated this study. The prevalence of diarrhoea among children from poor and non-poor households varied significantly and was nested at both the neighbourhood and country levels. We identified countries with pro-poor inequalities and those with pro-non-poor inequalities. There were unique variabilities in the factors that drive pro-poor inequalities in the development of diarrhoea across these countries. The findings from this article highlight the need for multiple approaches to understand and tackle the different factors that contributed to the inequalities in the risk of diarrhoea between the children from poor and non-poor households in LMICs. We found significant pro-poor inequality in 29 of all the 57 countries and pro-non-poor inequality in 7 of the countries, whereas there were insignificant gaps in the remaining countries.

The RDs in the prevalence of diarrhoea between children from poor and non-poor households showed that the fixed effect of pro-poor inequality was widest in Cameroon, whereas the fixed effect of pro-non-poor inequality was widest in Timor-Leste on the aggregate. These findings might be attributed to the difference in the socio-demographic, environmental and behavioural characteristics among the poor and non-poor households. Results of other studies agreed with this finding.[46–51] Diarrhoea was found to be more prevalent among infants than among those aged 12–59 months, irrespective of their households' poverty status. This finding is consistent with that of earlier studies.[52–55] The variations by the childrens' age may be ascribed to the fact that infants often have complementary feeding and higher exposure to contaminated food and water, which place them at higher risk of diarrhoea. This is an indication that greater efforts should be placed on prevention of diarrhoea among children especially at the earlier days of life.

Overall, the largest contributions to the pro-poor inequalities in having diarrhoea are maternal education, access to media, neighbourhood SES, place of residence, birth order and maternal age. We found an interesting pattern in the relationship and closeness among these compositional and contextual factors. The mothers' educational attainment and access to media were the greatest contributors to the inequalities and were clustered together, whereas the neighbourhood SES, rural-urban differences in the place of residence, birth order and maternal age formed another cluster. These two clusters later merged and formed a single cluster, which helped to explain the gaps in poor-non-poor diarrhoea prevalence.

The central role of maternal education in individuals' empowerment, well-being, access to quality information and capacity to make the right decisions cannot be overemphasised. Education, especially among women, is a gateway to opportunities, and it has been serially associated with health outcomes in the literature.[56–59] The significance of maternal education to wealth inequality as found in the present study has several implications. First, there is a need for most LMICs to develop and strengthen policies on the education of women as a means of family economic empowerment. Second, there is a dire need for public health policies, interventions and programmes that particularly inform and train mothers on how diarrhoea could be prevented. In addition, increasing the knowledge of mothers and all household members in general to join the diarrhoea prevention wagon is a must if diarrhoea prevalence should be drastically reduced.

Access to radio, television or newspaper can enhance mothers' knowledge about diarrhoea prevention practices. For instance, access to media remains the broader channel through which mothers could know that using Aquaguard could help prevent microbial contamination in water.[60] It suffices to say children whose mothers had media access as a result of having a form of education were less likely to develop diarrhoea than children whose mothers had not attended any formal education. This may be ascribed to the fact that education is likely to enhance household health and sanitation practices and also encourage behavioural changes at the household level.[46–51]

The neighbourhood SES, a composite measure of the community's proportion of women who are unemployed, illiterate and rural dwellers, was significant to pro-poor inequalities in having diarrhoea. A similar assertion has been made in earlier reports that the SES is the major driver of health outcomes in developing countries.[11,13,16–18] Therefore, concerted efforts are needed to ensure the overall community's SES through individuals' empowerment.

Rural-urban divides in the place of residence of children also contributed to the pro-poor inequality in the development of diarrhoea among U5C. The literature is replete that children who lived in rural areas coupled with lower means of livelihood are at higher odds of poorer health outcomes.[61] This could be as a result of limited economic capabilities, poor access to healthcare facilities and poor sanitation in rural areas. For instance, rural dwellers in countries such as Bangladesh, India, Malawi, Zambia and Peru and most sub-Saharan African countries, the source of drinking water is mainly from rivers, ponds and streams, which are prone to contamination.[62] In most rural areas, there are no improved toilet types, so open defecation prevails. Poor disposal of excreta is the main risk factor for diarrhoeal diseases.[4,20,63,64] More so, children with diarrhoeal disease may easily transmit the disease to others who live in the same area, especially in rural neighbourhoods with high poverty rates.

We found interesting results in the categorisation of countries by the distribution of the prevalence of diarrhoea and the RDs in having diarrhoea among children from poor and non-poor households. The categorisation includes countries with a high prevalence of diarrhoea and high pro-poor inequality such as Cameroon, Togo, Yemen, the Dominican Republic and Liberia and those with a low prevalence of diarrhoea and high pro-poor inequality such as South Africa, Rwanda, Nigeria and Tajikistan. Of particular concern is the group of countries with high prevalence and high pro-poor inequality. The variations across these countries were explainable by disparities in educational attainment, access to media, available country-level policies and programmes for child health, as well as political and economic instability. There is a need for countries with a high prevalence of diarrhoea and high pro-poor inequality to take a cue from what is been done right in the countries with low prevalence and low pro-non-poor inequalities.

Strengths and limitations

Secondary data were used for the analysis. The data required that mothers should recall a recent episode of diarrhoea without any means of verification by the interviewers. Besides, correct identification of what diarrhoea is could be a potential recall bias. Data analysis of three-quarters of a million children spread across 57 LMICs is a major strength of our study as it showed a wide coverage and generalisability. We quantified the magnitude of the

factors associated with pro-poor inequalities in the development of diarrhoea using the Fairlie decomposition methods that provided robust evidence of wealth-related inequalities after controlling for the exposure variables.

Conclusions

Diarrhoea remains a major challenge in the majority of the LMICs with a wide range of pro-poor inequalities. These disparities were explained by compositional and contextual factors that cut across individual-, household- and community-level factors. The overall significance of our determinate variable in explaining the difference in diarrhoea prevalence is a pointer to the fact that empowerment of individuals is very important to achieving favourable child health outcomes in most countries. The magnitude of the contributions of factors associated with the pro-poor inequalities varied widely across the countries. Thus, multifaceted geographically specific intervention may prove to be a potent approach to address the poor and non-poor differentials in the risk of diarrhoea among U5C, with policies tailored to country-specific conditions.

Author statements

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Ethical approval

This study was based on the analysis openly available on secondary data. The Institutional Review Board (IRB) of ICF Macro at Fairfax, Virginia, in the USA reviewed and approved the MEASURE Demographic and Health Surveys Project Phase III. The 2010–2018 Demographic and Health Surveys are categorised under that approval. The IRB of ICF Macro complied with the United States Department of Health and Human Services requirements for the 'Protection of Human Subjects' (45 CFR 46). Written informed consent was obtained from every study participant before participation, and all information was collected without identifiers and kept confidentially. ICF Macro permitted the authors to use the data. The full details of the ethical approvals can be found at http://dhsprogram.com.

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Availability of data and materials

The data supporting this article is available at http://dhsprogram.com.

Author contributions

A.F.F. conceptualised, designed the study, curated and analysed the data. A.F.F., O.O.P., E.K.A., O.S.F. and O.A.U. contributed to the literature search, figures, data interpretation and writing of the manuscript.

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Original Research

Evaluation of work resumption strategies after COVID-19 reopening in the Chinese city of Shenzhen: a mathematical modeling study

Lu Bai [a, i], Haonan Lu [b, i], Hailin Hu [b], M. Kumi Smith [c], Katherine Harripersaud [d], Veronika Lipkova [e], Yujin Wen [b], Xiuyan Guo [b], Wei Peng [b], Chenwei Liu [b], Mingwang Shen [a], Alfred Chixiong Shen [b], Lei Zhang [a, f, g, h, *]

[a] China-Australia Joint Research Center for Infectious Diseases, School of Public Health, Xi'an Jiaotong University Health Science Center, Xi'an, Shaanxi, 710061, PR China
[b] AI Application Research Center, Huawei Technologies Co., Ltd. Shenzhen, Guangdong, 518000, PR China
[c] Division of Epidemiology and Community Health, University of Minnesota Twin Cities, Twin Cities, United States
[d] ICAP, Mailman School of Public Health, Columbia University, New York, United States
[e] Medical Sciences Division, University of Oxford, Oxford, United Kingdom
[f] Melbourne Sexual Health Centre, Alfred Health, Melbourne, Australia
[g] Central Clinical School, Faculty of Medicine, Nursing and Health Sciences, Monash University, Melbourne, VIC, Australia
[h] Department of Epidemiology and Biostatistics, College of Public Health, Zhengzhou University, Zhengzhou 450001, Henan, China

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Work resumption
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ABSTRACT

Objectives: As China is facing a potential second wave of the epidemic, we reviewed and evaluated the intervention measures implemented in a major metropolitan city, Shenzhen, during the early phase of Wuhan lockdown.

Study design: Based on the classic SEITR model and combined with population mobility, a compartmental model was constructed to simulate the transmission of COVID-19 and disease progression in the Shenzhen population.

Methods: Based on published epidemiological data on COVID-19 and population mobility data from Baidu Qianxi, we constructed a compartmental model to evaluate the impact of work and traffic resumption on the epidemic in Shenzhen in various scenarios.

Results: Imported cases account for most (58.6%) of the early reported cases in Shenzhen. We demonstrated that with strict inflow population control and a high level of mask usage after work resumption, various resumptions resulted in only an insignificant difference in the number of cumulative infections. Shenzhen may experience this second wave of infections approximately two weeks after the traffic resumption if the incidence risk in Hubei is high at the moment of resumption.

Conclusion: Regardless of the work resumption strategy adopted in Shenzhen, the risk of a resurgence of COVID-19 after its reopening was limited. The strict control of imported cases and extensive use of facial masks play a key role in COVID-19 prevention.

Introduction

The coronavirus SARS-COV-2 pandemic had led to more than 31 million infections, and over 960,000 individuals died of the coronavirus disease 2019 (COVID-19), as of September 21st, 2020.[1] The COVID-19 epidemic broke out in early December 2019 in Wuhan, the provincial capital city of Hubei in China. The virus proved to be capable of interpersonal transmission even in the absence of overt symptoms, which, in combination with increased travel before the Lunar New Year, resulted in its rapid spread to all 31 Chinese provinces.[2–5] To curb the epidemic, the Wuhan authority imposed a strict metropolitan-wide 'lockdown' of Wuhan on Jan 23rd. During the 76 days (Jan 23rd to Apr 8th) lockdown, the operation of all public transportation in Wuhan was suspended, and all airport and railway stations were also temporarily closed.[6] Within two days of the Wuhan lockdown, all 31 Chinese provinces and regions have launched the 'level 1 public health emergency response', which is the highest level of state of emergency.[7] The lockdown has

* Corresponding author. Department of Epidemiology and Biostatistics, School of Public Health, Xi'an Jiaotong University Health Science Center, Xi'an, Shaanxi, 710061, PR China.
E-mail address: lei.zhang1@xjtu.edu.cn (L. Zhang).
[i] These authors contributed equally.

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effectively limited the movement of the population and reduced the speed of spread of COVID-19 outside Hubei.[2,9–10] Since the reopening of Wuhan city, the COVID-19 epidemic in China had been largely brought under control. As of September 21st, 2020, a total 85,291 cases were reported, and 4634 died of COVID-19 in Mainland China.[1] However, multiple small outbreaks have been reported across the country over the past three months, leading to a partial lockdown of the affected areas and vast cancellation of flights. As China is facing a potential second wave of the epidemic, we reviewed the intervention measures implemented in a major metropolitan city, Shenzhen, during the early phase of Wuhan lockdown. The past experiences may provide us with insights for future COVID-19 control and prevention.

Shenzhen was a megacity dominated by 8.5 million domestic migrants. This population accounted for 65% of its residents and formed the main labor force for Shenzhen's economy.[11] Before the implementation of 'level 1' response on January 23rd, Shenzhen's population outflow just peaked, with the total population outflow exceeding 9.5 million. Indeed, Shenzhen is a large city with high population mobility: during the period from January 1st to February 14th, 2020, the total population inflow to Shenzhen exceeded 8.4 million.[12] Because asymptomatic individuals can be infectious during the incubation period,[5,13–15] the return of domestic migrants may serve as a potential source of new infections. Hence, it is possible that the population influx from other parts of China, especially Hubei, may have a significant impact on the epidemic in Shenzhen in the early stages after work resumption.[16]

Taking into consideration the high population mobility, we constructed a compartmental model to simulate the transmission of COVID-19 and disease progression in the Shenzhen population. Based on which, we aimed to evaluate the trend of the COVID-19 epidemic for various scenarios of work resumption strategies for the returning residents in Shenzhen.

Methods

Data sources

We collected early published epidemic data on COVID-19 cases in Shenzhen, which were obtained from the open data platform of Shenzhen Municipal Government.[17] The population mobility data were retrieved from Baidu Qianxi with location-based services having nearly 9 billion location requests each day,[18] which was also in the public domain.[12]

Model description

Definition of disease stages

Based on the classical epidemiological dynamic SEITR model,[8,19] we proposed a M-SEITR model to evaluate the development of the epidemic, where 'M' stands for in-time population mobility correction (Fig. 1).[20] In the M-SEITR model, the population was divided into five compartments, which include susceptible individuals (S), individuals during the incubation period (E), infected but undiagnosed individuals (I), diagnosed individuals with treatment (T), recovered individuals (R), and death individuals (D). The total population size was denoted as N, ($N=S+E+I+T+R$).

The transmission of COVID-19 in the population

The schematic disease progression diagram is demonstrated in Fig. 1 (details in the Appendix). The model took into account the effects of facial mask usage $p(t)$ and interpersonal contact m per day on the COVID-19 epidemics. Specifically, we used a multinomial distribution to describe the transmission probability caused by interpersonal contact, which depends on the number of daily interpersonal contacts and the probability of transmission per contact (β). Comparing with elsewhere in the world, the Chinese Government had developed guidelines for the use of masks and enforced a more strict facial mask-wearing practice, especially in public places and on public transports.[8,21] At the initiation of the simulation at January 1st, where there were no confirmed cases reported in Shenzhen, we assumed the background facial mask rate to be zero (Fig. 1).

Impact of work resumption

We assumed that the probability of transmission decreased with the reduction of interpersonal contact and the increase in the use of facial masks. We assumed that the vast majority of citizens would maintain the habit of wearing masks until the end of the epidemic (even after work resumption). Furthermore, work resumption would increase the frequency of interpersonal contacts, which may further affect the trajectory of the epidemic. Importantly, we assumed that contact frequency m after work resumption would increase three-fold relative to the frequency of contact with family members before work resumption.[22] And the various resumption of work ratio at different dates in the resumption strategies below affects the population mobility in Shenzhen. With the increase in the resumption of work ratio, the returning population in Shenzhen also increased, including people at different stages of disease (Appendix).

Simulation of population mobility

For population mobility, we assumed the population would return to Shenzhen after resumption in the same size and speed as they left the city before the strict control was implemented. We assumed that population mobility could affect three subpopulations, the susceptible individuals (S), the asymptomatic latent individuals (E), and undiagnosed infected individuals (I) (Fig. 1). The parameters and mathematical formulation for population mobility were listed in the Appendix. Imported cases first appeared in Shenzhen on January 4th, and the first local case of public transmission occurred on January 15th, considering the estimated incubation period of COVID-19 (3–7 days),[11,15,23,24] we conservatively regard January 1st as the starting point of the Shenzhen epidemic.

Scenarios for evaluation

The epidemic situation in Hubei may impact on Shenzhen in two ways. First, because the number of confirmed cases in Hubei has increased substantially on February 12th because of the change of diagnostic criteria and the progress of patient admission,[25] it is likely that the number of latent infections among the Hubei travelers to Shenzhen in January might have been underestimated. The extent of control of the imported cases in Shenzhen in late January would impact substantially on the epidemic in Shenzhen. Second, the inflow of asymptomatic infections to Shenzhen after work resumption may be affected by the epidemic situation in Hubei.

We created four scenarios to reflect the potential intervention status in Shenzhen. Scenario 1 represents a prompt control of the inflow of the infected population from Hubei into Shenzhen in January and a low incidence risk in Hubei in March after work resumption. Scenario 2 represents a prompt control of the inflow of the infected population from Hubei in Shenzhen in January but a high incidence risk in Hubei in March after work resumption. Scenario 3 represents a delayed control of the inflow of the infected population from Hubei in Shenzhen in January and a low incidence

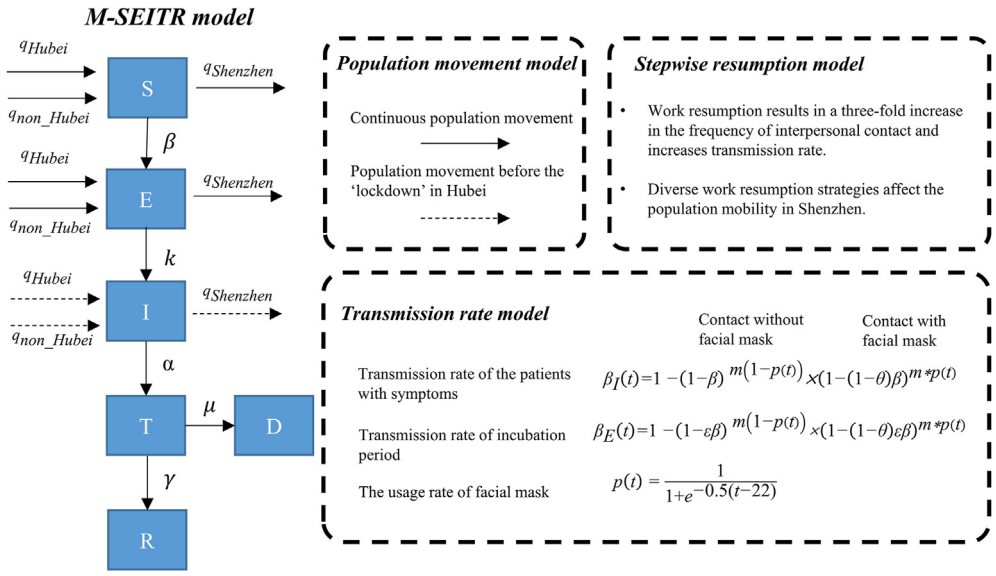

Fig. 1. Schematic diagram of M-SEITR model. On the basis of SEITR model, the population migration, stepwise resumption of work strategies, and the transmission rate is simulated in detail. In the process of population movement, the model took into account the inflow to Shenzhen of Hubei travelers and non-Hubei travelers and the population outflow from Shenzhen, as well as the changing effect of the total population of Shenzhen at the same time. The transmission rate model combines the changes in the average number of interpersonal contacts per day and the effects of the facial mask usage.

risk in Hubei in March after work resumption. Scenario 4 represents a delayed control of the inflow of the infected population from Hubei in Shenzhen in January but a high incidence risk in Hubei in March after work resumption. The high and low incidence risk refers to the simulation of the rate of infection in Hubei with various level of decline in the epidemic trend.

Resumption strategies

To evaluate the possible impact of work resumption on the epidemic, we identified six stepwise resumption s in each scenario. These included (1) full resumption of work from February 10th; (2) scheme 1, a partial resumption of 57% on February 10th followed by a full resumption on February 17th; (3) scheme 2, a partial resumption of 51% on February 10th followed by a full resumption on February 17th; (4) scheme 3, a partial resumption of 51% on February 10th, then 63% on February 17th, followed by a full resumption on February 24th; (5) scheme 4, an increasing partial resumption of 39%, 51%, and 63% on February 10th, 17th, and 24th, respectively, followed by a full resumption on March 2nd; (6) scheme 5, a partial resumption of 57% and 74% on February 10th and 17th, respectively, followed by a full resumption on February 24th. The calculation of the partial resumption is based on the type of industry, immediate urgency for the resumption and their impact on the spread of the epidemic. In general, industries related to people's daily necessities were prioritized. These were followed by industries that were essential but allowed for 'work from home', then industries that may be resumed in the near future, and those can be further delayed. The full explanation of the resumption schemes was listed in the Appendix (Tables S1 and S2).

Model calibration

We calibrated the model parameters based on the of confirmed cases of COVID-19 published in by Shenzhen Center for Disease Control (Appendix). Overall, the calibrated model demonstrated good consistency between the model output and the reported number of imported cases.

Results

Of 406 confirmed cases that were reported in Shenzhen, 238 cases (58.6%) were imported. Of these imported cases, 153 cases (37.7%) were from Hubei. There were 105 local cases due to household transmission and 63 due to public contacts, accounting for 25.9% and 15.5% of all reported cases. Table 1 demonstrated the composition of COVID-19 cases in Shenzhen.

We predicted the cumulative number of infected cases for the six resumption strategies based on the four intervention scenarios in Shenzhen. When a prompt control of the inflow of the infected population was in place, and incidence risk in Hubei was low (scenario 1), full work resumption from February 10th would result in 68 additional infected cases between February 10th and April 30th, and the cumulative infected cases would reach 456 (453–458) by April 30th. For the other five stepwise resumption schemes in scenario 1, the cumulative number of infected individuals was reduced compared with that of full work resumption scheme, but the difference was small (3–5 fewer cases by April 30th). By contrast, when a prompt control of inflow of infected population was in place but the incidence risk in Hubei was high (scenario 2), the number of cumulative infected cases would reach 542 (540–544) in the event of full work resumption. However, if the control of the infected population from Hubei in Shenzhen in January was delayed, full resumption of work would result in a much higher number of cases by April 30th (scenario 3, low incidence risk in Hubei: 922 [848–995]; scenario 4, high incidence risk in Hubei: 1044 [936–1153]). In scenarios 2, 3, and 4, the differences between work resumption schemes were small, and by the end of April, the cumulative number of infected cases only differed by 2–4, 41–73, and 54–99, respectively (Table S4).

The estimated number of individuals who were infected but undiagnosed demonstrated a similar trend across all four scenarios, reaching a peak (98–158 cases) around the end of January, before gradually declining. The traffic resumption in Hubei province may lead to a second but significantly smaller peak (24–26 cases) if Hubei remains a high incidence risk in March. After the second peak, the trend would continue to decline to zero (Fig. 2).

Table 1
The number of reported cases of COVID-19 in Shenzhen 3 weeks after Wuhan's lockdown.

Case type		The number of cases	Proportion
Imported cases[a]	Hubei travelers	153	37.7%
	Non-Hubei travelers	85	20.9%
Local household transmission		105	25.9%
Local public transmission		63	15.5%
Total		406	–

[a] If a family group arrived Shenzhen and more than one member was diagnosed positive, then only one case was regarded as 'imported case' and the rest were local household transmission cases.

Discussion

Our study demonstrated that imported cases account for most (58.6%) of all reported cases in Shenzhen. In particular, imported cases from Hubei account for 37.7%. If Shenzhen maintains strict control measures with regard to the inflow population, and its citizens maintain a high level of mask usage even after the resumption of operations, the epidemic will gradually subside, with few differences between the proposed resumption schemes. If intercity travel is restored when Hubei still has a high incidence risk, Shenzhen may experience a second wave of infections.

Our analysis indicates that the COVID-19 epidemic in Shenzhen would mainly result from imported cases and household transmission, with the local public transmission being relatively limited.[26] Notably, only one-quarter of the cases were due to household transmission, which stands in sharp contrast to the 56–61% in Hubei province.[1,27] As the virus is highly contagious and protective measures in a household setting are usually limited, the chance of transmission due to an asymptomatic infected household member is very high. The low percentage of household transmission indicates that early public health measures in Shenzhen have been effective. In particular, strict temperature monitoring, timely isolation, contact tracing and treatment for confirmed cases seems to have played a major role.[28]

We found that different work resumption strategies have little impact on the overall trajectory of the epidemic in Shenzhen. This may be for a number of reasons. First, because the number of undiagnosed infected cases in Shenzhen was small and the epidemic was well controlled in its early phase, the impact of various resumption strategies makes little difference to the epidemic.

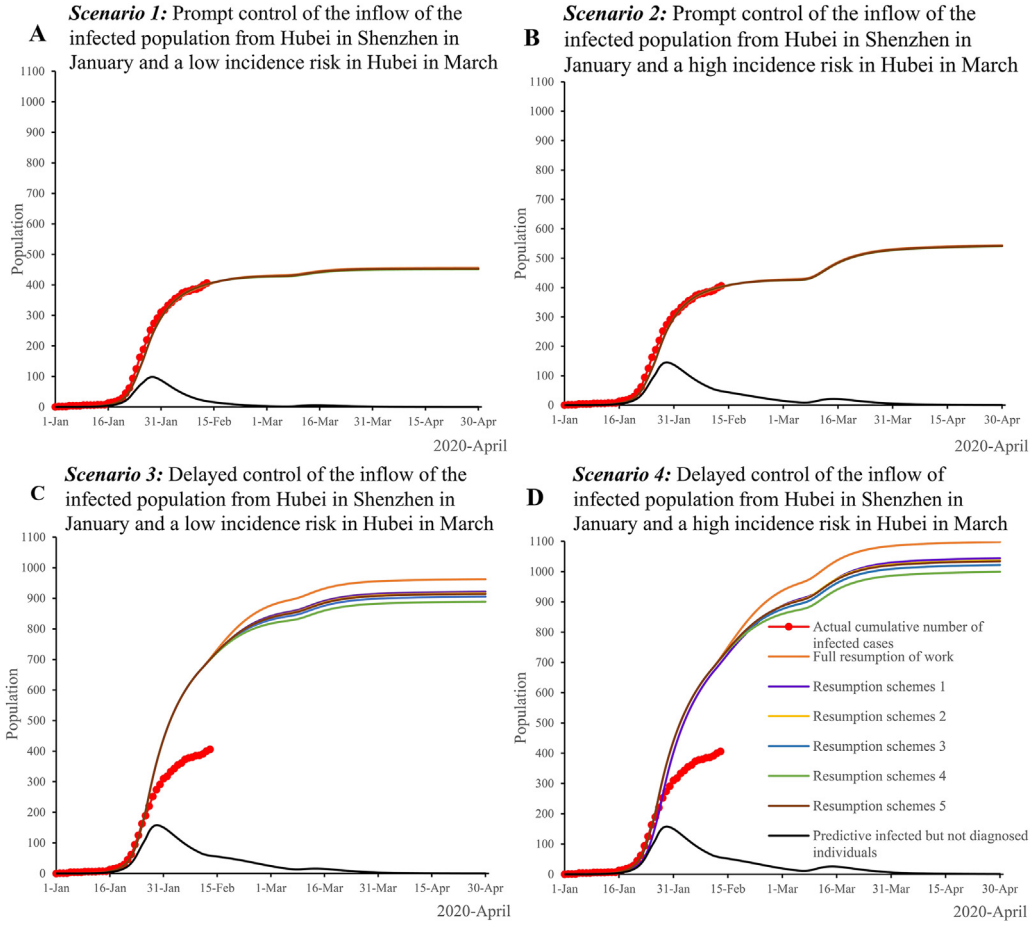

Fig. 2. Under the four scenarios of the epidemic hypothesis of Shenzhen and the six resumption s, the changing trend of the predictive cumulative number of infected cases (*C*) and the predicted number of infected but undiagnosed individuals (*I*) in Shenzhen. (1) Full resumption of work from February 10th; (2) 1, a partial resumption of 57% on February10th followed by a full resumption on February 17th; (3) 2, a partial resumption of 51% on February 10th followed by a full resumption on February 17th; (4) 3, a partial resumption of 51% on February 10th, then 63% on February 17th, followed by a full resumption on February 24th; (5) 4, an increasing partial resumption of 39%, 51% and 63% on February 10th, 17th, and 24th, respectively, followed by a full resumption on March 2nd; (6) 5, a partial resumption of 57% and 74% on February 10th and 17th, respectively, followed by a full resumption on February 24th.

Second, as facial masks were widely used, including asymptomatic individuals, an increase in the frequency of interpersonal contacts caused by work resumption does not effectively increase the transmission of SARS-CoV-2, suggesting that the ongoing personal protective measures were crucial to the process of the city reopening.[29]

Our analysis showed that if Hubei had restored traffic in early March, Shenzhen might have experienced the second wave of the outbreak at a later point that month. However, the number of imported cases is small, and the threat is limited. The Shenzhen government has imposed strict resumption strategies that encourage business to implement altered off-peak dining, such as reducing the frequency and scale of meetings and minimizing staff gatherings.[28] These measures are key in preventing a second outbreak in Shenzhen.

This study has several limitations. First, the model did not take into account the spread caused by the use of public transportation (e.g. subway and buses); consequently, the risk of transmission in public spaces may have been underestimated. Second, we modeled the population mobility model based on data from Baidu Qianxi, which may not fully account for the actual movement of the population. Third, our model did not take into consideration of overseas imported cases. Besides, we did not take into account the human behaviors (e.g. social distancing and hands washing) and may overestimate the COVID-19 epidemic in Shenzhen. Yet this allows our evaluation of prevention measures to be more conservative.

In conclusion, regardless of the work resumption strategy adopted in Shenzhen, the risk of a resurgence of COVID-19 after its reopening was limited. The strict control of imported cases and extensive use of facial masks play a key role in COVID-19 prevention.

Author statements

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Ethical approval

Not applicable, as no patient personal information is involved in this study.

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Competing interests

All authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this article.

Availability of data and materials

The data that support the findings of this study are available from the open data platform of Shenzhen Municipal Government[17] and the population mobility data were retrieved from Baidu Qianxi,[18] which was also in the public domain[12].

Disclaimer

The funding agencies had no involvement in the design and conduct of the study; collection, management, analysis, and interpretation of the data; preparation, review, or approval of the manuscript; or decision to submit the manuscript for publication.

Authors' contributions

HL, HH, MS, ACS, and LZ conceived and designed the study. HL, HH, YW, XG, WP, and CL analyzed the data, carried out the analysis, and performed numerical simulations. LB wrote the first draft of the manuscript. MKS, KH, VL, and YW critically read and revised the manuscript. All authors contributed to writing the article and agreed with manuscript results and conclusions.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.puhe.2020.12.018.

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Short Communication

Healthcare indicators associated with COVID-19 death rates in the European Union

C. Mattiuzzi [a], G. Lippi [b, *], B.M. Henry [c]

[a] Provincial Agency for Social and Sanitary Services, Trento, Italy
[b] Section of Clinical Biochemistry, University of Verona, Verona, Italy
[c] Cardiac Intensive Care Unit, The Heart Institute, Cincinnati Children's Hospital Medical Center, Ohio, USA

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ABSTRACT

Objectives: Identification of environmental and hospital indicators that may influence coronavirus disease 2019 (COVID-19) mortality in different countries is essential for better management of this infectious disease.

Study design: Correlation analysis between healthcare system indicators and COVID-19 mortality rate in Europe.

Methods: For each country in the European Union (EU), the date of the first diagnosed case and the crude death rate for COVID-19 were retrieved from the John Hopkins University website. These data were then combined with environmental, hospital and clinical indicators extracted from the European Health Information Gateway of the World Health Organization.

Results: The COVID-19 death rate in EU countries (mean 1.9 ± 0.8%) was inversely associated with the number of available general hospitals, physicians and nurses. Significant positive associations were also found with the rate of acute care bed occupancy, as well as with the proportion of population who were aged older than 65 years, overweight or who had cancer. Total healthcare expenditure, public sector health expenditure and the number of hospital and acute care beds did not influence COVID-19 death rate.

Conclusions: Some common healthcare system inadequacies, such as limited numbers of general hospitals, physicians and nurses, in addition to high acute care bed occupancy, may be significant drivers of nationwide COVID-19 mortality rates in EU countries.

Although coronavirus disease 2019 (COVID-19) has spread around the world, there is a broad divergence in terms of COVID-19 mortality across different countries, as recently highlighted by Teixeira da Silva and Tsigaris.[1] In addition to identifying clinical and laboratory predictors of individual disease progression, it is also vital to recognise that environmental and hospital indicators may influence the impact of COVID-19, thus, in part, explaining the wide heterogeneity of death rates observed across different countries.

To investigate this matter further, the present study retrieved the date of the first diagnosed case and the crude death rate for COVID-19 for each country within the European Union (EU) from the John Hopkins University website.[2] These data were then combined with a number of environmental, hospital and clinical indicators extracted from the European Health Information Gateway (EHIG) of the World Health Organization (WHO).[3] More specifically, separate queries were made in the EHIG database, using the specific keywords (i.e. healthcare indicators) listed in Table 1, to retrieve data for each EU country. According to the WHO regional office for Europe, the information contained in the EHIG repository is derived from various reliable sources, including WHO/Europe's technical programmes and partner organisations, such as Eurostat, the Organisation for Economic Co-operation and Development and the United Nations.[3] The most recent available data of the EU healthcare indicators were imported into a Microsoft Excel file (Microsoft, Redmond, WA, United States) along with the country-specific COVID-19 crude death rates. After logarithmic data conversion, a multiple linear regression analysis was carried out to identify potentially independent associations; death rate (%) was set as the dependent variable, whilst environmental, healthcare and clinical indicators were

* Corresponding author. Section of Clinical Biochemistry, University Hospital of Verona, Piazzale L.A. Scuro, 10, 37134, Verona, Italy. Tel.: +39 045 8122970; fax: +39 045 8124308.
E-mail address: giuseppe.lippi@univr.it (G. Lippi).

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Table 1
Association between environmental, hospital and clinical indicators with coronavirus disease 2019 (COVID-19) mortality rate in European Union countries.

Parameter	β coefficient (95% CI)	*P*-value
1st diagnosed case (date)	−284 (−1002 to 433)	0.380
Environmental indicators		
Mean yearly temperature (°C)	0.274 (−0.484 to 1.031)	0.421
Density (population/km^2)	−0.243 (−0.493 to 0.007)	0.055
Proportion of urban population (%)	−1.190 (−3.970 to 1.590)	0.345
People per room in occupied housing unit (number)	−0.384 (−1.595 to 0.826)	0.477
Gross domestic product (US$ per capita)	0.722 (−0.003 to 1.447)	0.051
Hospital indicators		
Total healthcare expenditure (US$ per capita)	−0.002 (−1.597 to 1.594)	0.998
Public sector healthcare expenditure (% of total health expenditure)	−0.737 (−1.737 to 0.264)	0.125
General hospitals (per 100,000)	−0.513 (−0.918 to −0.107)	**0.020**
Hospital beds (per 100,000)	0.037 (−0.786 to 0.860)	0.918
Acute care beds (per 100,000)	1.039 (−0.194 to 2.273)	0.056
Acute care bed occupancy (%)	3.639 (1.743–5.534)	**0.003**
Physicians (per 100,000)	−1.494 (−2.792 to −0.196)	**0.039**
Nurses (per 100,000)	−1.290 (−2.242 to −0.339)	**0.015**
Clinical indicators		
Estimated life expectancy at birth (years)	−9.036 (−19.270 to 1.198)	0.075
Population aged >65 years (%)	3.019 (0.448–5.590)	**0.027**
Age-standardised current tobacco smoking in people >15 years (%)	−0.943 (−2.817 to 0.930)	0.273
Age-standardised overweight in people >18 years (%)	6.886 (0.347–13.426)	**0.042**
Incidence of cancer (per 100,000)	0.577 (0.117–1.037)	**0.041**
People self-assessing health as good (%)	2.170 (0.797–3.543)	**0.007**

CI, confidence interval. Statistically significant associations are given in bold.

set as independent variables. Statistical analyses was carried out using Analyse-it (Analyse-it Software Ltd, Leeds, UK), with significance set at $P < 0.05$. The analyses were based on electronic searches in unrestricted, publicly available databases, and therefore, no informed consent or ethical committee approval was required.

The results of this investigation are summarised in Table 1. The COVID-19 death rate in EU countries (mean 1.9 ± 0.8%) varied between 0.6% in Cyprus and 3.6% in Bulgaria, and it was inversely associated with the number of available general hospitals, physicians and nurses. Significant positive associations with mortality were found with the rate of acute care bed occupancy, as well as with the proportion of the population who were aged >65 years, overweight or who had cancer. A positive association with mortality was also found with the proportion of the population self-assessing their health as good. Importantly, neither total healthcare expenditure, public sector health expenditure nor the number of hospital and acute care beds were found to influence COVID-19 mortality rate. Moreover, in the present analysis, no environmental parameters were found to have a significant influence on COVID-19 mortality (Table 1), although the association with gross domestic product per capita and population density were of borderline statistical significance.

Taken together, the results of this study suggest that some common healthcare system inadequacies, such as a limited number of general hospitals, physicians and nurses, along with high acute care bed occupancy, may be significant drivers of nationwide COVID-19 mortality rates in EU countries. Additional parameters that were found to be associated with increased COVID-19 death rate included, as expected, a high proportion of population aged >65 years, along with a high national burden of overweight individuals and cancer diagnoses. This is not surprising because these parameters have been repeatedly shown to individually contribute to a poor prognosis in COVID-19.[4] Notably, no significant associations of COVID-19 mortality were found with total healthcare expenditure, public sector health expenditure or availability of hospital and acute care beds.

It is now unquestionable that COVID-19 has imposed a remarkable burden on healthcare resources around the world, with significant concerns over the capacity to manage the huge number of COVID-19 cases that are diagnosed each day. According to this analysis, it seems that is not the total amount of money spent by national governments but rather the way this money is spent on healthcare and hospital organisation that may have the most significant influence on COVID-19 management and outcomes. In fact, it seems that even a large availability of hospital or acute care beds may be ineffective in reducing COVID-19 mortality if this is not combined with increased availability of physicians and nurses and improved hospital accessibility.

Author statements

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Original Research

Public health information on COVID-19 for international travellers: lessons learned from a mixed-method evaluation

T. Zhang [a], C. Robin [b, c, d, e], S. Cai [a], C. Sawyer [f, g], W. Rice [b], L.E. Smith [h, i], R. Amlôt [c, i, j], G.J. Rubin [h, i], R. Reynolds [a, c], L. Yardley [c, k, l], M. Hickman [a, c], I. Oliver [a, b, c], H. Lambert [a, c, *]

[a] Population Health Sciences, Bristol Medical School, University of Bristol, Bristol, UK
[b] Field Epidemiology, Field Service, National Infection Service, Public Health England, Bristol, UK
[c] NIHR Health Protection Research Unit in Behavioural Science and Evaluation, Bristol Medical School, University of Bristol, Bristol, UK
[d] NIHR Health Protection Research Unit in Emerging and Zoonotic Infections, University of Liverpool, Liverpool, UK
[e] NIHR Health Protection Research Unit in Gastrointestinal Infections, University of Liverpool, Liverpool, UK
[f] UK Field Epidemiology Training Programme, Global Public Health Division, Public Health England, London, UK
[g] Communicable Disease Surveillance Centre, Public Health Wales, Cardiff, UK
[h] Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, UK
[i] NIHR Health Protection Research Unit in Emergency Preparedness and Response, King's College London, London, UK
[j] Behavioural Science Team, Emergency Response Department Science and Technology, Public Health England, UK
[k] School of Psychological Sciences, University of Bristol, Bristol, UK
[l] Department of Psychology, University of Southampton, Southampton, UK

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ABSTRACT

Objectives: In the containment phase of the response to the COVID-19 outbreak, Public Health England (PHE) delivered advice to travellers arriving at major UK ports. We aimed to rapidly evaluate the impact and effectiveness of these communication materials for passengers in the early stages of the pandemic.
Study design: The study design used is the mixed-methods evaluation.
Methods: A questionnaire survey and follow-up interviews with passengers arriving at London Heathrow Airport on scheduled flights from China and Singapore. The survey assessed passengers' knowledge of symptoms, actions to take, and attitudes towards PHE COVID-19 public health information; interviews explored their views of official public health information and self-isolation.
Results: One hundred and twenty-one passengers participated in the survey and 15 in follow-up interviews. Eighty three percentage of surveyed passengers correctly identified all three COVID-19 associated symptoms listed in PHE information at that time. Most could identify the recommended actions and found the advice understandable and trustworthy. Interviews revealed that passengers shared concerns about the lack of wider official action, and that passengers' knowledge had been acquired elsewhere as much from PHE. Respondents also noted their own agency in choosing to self-isolate, partially as a self-protective measure.
Conclusion: PHE COVID-19 public health information was perceived as clear and acceptable, but we found that passengers acquired knowledge from various sources and they saw the provision of information alone on arrival as an insufficient official response. Our study provides fresh insights into the importance of taking greater account of diverse information sources and of the need for public assurance in creating public health information materials to address global health threats.

* Corresponding author. Population Health Sciences, Bristol Medical School, University of Bristol, Bristol, UK.Tel.: +44(0)117 9096406.
E-mail address: h.lambert@bristol.ac.uk (H. Lambert).

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Introduction

With international arrivals growing to 1.186 billion in 2015,[1] increasing global connectivity has increased pressure on cross-national prevention and containment of disease outbreaks, including the COVID-19 pandemic. Recent studies show a central role of travel in the spread of COVID-19, with evidence of a strong correlation between domestic travellers departing from Wuhan and the subsequent seeding of COVID-19 epidemics in their arrival cities.[2] Internationally, the countries receiving the largest traveller volumes from Wuhan, such as Thailand and Japan, also confirmed the highest COVID-19 cases outside China in January 2020,[3] along with certain in-flight COVID-19 transmission cases reported worldwide.[4] The first cases of COVID-19 in England were reported on 29th January in two recently arrived travellers from China. Initial cases were mostly associated with international travel.

The ongoing risk associated with travel highlights the importance of interventions that target arriving passengers to control transmission and protect the public. During the containment phase of the UK's COVID-19 response, whereas the outbreak epicentre was in Asia, public health information was delivered to travellers arriving at UK ports (summarised in Box 1).

Box 1
Summary of measures at UK ports for arriving travellers since containment phase of the COVID-19 response.

- The Airport Public Health Monitoring Operations Centre established by Public Health England (PHE) was activated on 25th January to monitor all direct flights from China to LHR, and operations were extended to include all direct flights to London Gatwick and Manchester on 29th January until travel restrictions were implemented.
- Measures directed at passengers travelling from affected countries into the UK included a broadcast message to passengers made on incoming aircraft, to encourage travellers to report their illness; posters containing COVID-19–related public health advice displayed at these three airports; and leaflets containing this advice provided to passengers by airlines on board the flight and/or made available on arrival.
- Contact tracing was undertaken when a case was reporting including flights and other transport.
- Since 8th June, people entering or return to the UK are required to provide their journey and contact details and self-isolate for 14 days if arriving from an affected country, with penalties of up to £1000 for breaking this rule.[5] These regulations continue to be amended, with exemptions for travellers arriving from specified countries of origin.

Provision of public health advice at ports of entry was last used in the UK in during the 2014/2015 Ebola outbreak in West Africa, and travellers considered this reassuring.[6] Emerging viral diseases, such as Ebola, have caused widespread panic and travel warnings; however, COVID-19 has more serious impact on travel medicine and tourism industry than Ebola and other public health emergencies of international concern.[7] This study aimed to evaluate the effectiveness and impact of Public Health England (PHE) COVID-19 communication materials (see Supplementary documents) for passengers arriving at UK airports during the containment phase of the response (24th January-12th March). The study was conducted at the request of the Department of Health and Social Care via the National Institute of Health Research. Adjustments to the study protocol were made due to the fast-changing situation as the number of flights carrying passengers into the UK dropped substantially in the monitoring period, from 16 to 18 flights daily from China (including Hong Kong) into London Heathrow (LHR) in the third week of January to nine flights per week by the end of January, reducing further in subsequent months. Internal LHR data indicate that in March, 123 flights arrived from China, Hong Kong and Singapore, one-fifth of the number in February.

Methods

We undertook a two-stage mixed-methods evaluation, starting with patient and public involvement interviews with Chinese students and staff at two UK universities returned to the UK from China in January and February 2020 (stage I), followed by a survey and semistructured interviews with air passengers returning to the UK from COVID-19 affected countries (stage II). The questionnaire and interview topic guides developed for stage II were based on stage I results. This article only reports findings from stage II.

Study population

Returning travellers aged 18 years and older from any nationality, arriving into LHR airport from affected countries after PHE leaflets and posters began to be distributed on 23rd January.

Sampling and methods

Cross-sectional survey

Passengers arriving at LHR airport on three scheduled flights on 4th March from Singapore and on 12th March and 13th March from China were recruited into the survey. PHE listed both countries as places of origin necessitating advice for travellers, with Hubei and Wuhan in China highlighted as requiring separate advice. Paper questionnaires in English, Mandarin and Cantonese, along with the PHE leaflets in English and simplified Chinese script, were issued by airline crew (who were given instructions in advance) to all passengers for completion before disembarkation. The short questionnaire collected information on: participants' knowledge of COVID-19 symptoms (Q1) and help-seeking behaviours (Q2); whether participants received the public health advice (Q3) and views on it (Q4); and demographic information (Q5-Q11).

Respondents were also invited to record their name and contact details if willing to take part in follow-up interviews. Researchers then met passengers at disembarkation points at LHR airport to collect completed questionnaires and consent passengers to follow-up interviews.

Semi-structured interviews

Passengers consenting to interview were contacted by email to confirm an interview time and language preference (English/Mandarin). After confirmation, one-to-one telephone interviews of approximately 30 min were conducted between 2nd-23rd April 2020.

During interviews, participants were asked about the COVID-19 information they received during their journey and their thoughts on the PHE information provided. Interviewees who reported having developed symptoms since arriving in the UK and had self-isolated were also asked about their views and experiences of self-isolation, using a separate topic guide.

All interviews were audio-recorded, and researchers created summaries of each interview. English interviews were transcribed *verbatim*; Mandarin interviews were transcribed directly into English.

Data analysis

Categorical data were described as proportions and continuous data as median with interquartile range (IQR). All analyses were conducted in Stata v15.1 (2017, StataCorp LLC, College Station, TX).

Interview transcripts were coded using open coding. An initial coding framework was collaboratively developed by four researchers (T.Z., S.C., C.S., W.R.) each coding one interview that they had conducted. Two (T.Z., S.C.) of the research team used the coding framework to index each transcript in NVivo 12 Pro. Coding was performed iteratively within and between transcripts; common categories emerged across the transcripts, indicating data saturation.[8]

Results

Survey results

Demographic characteristics

One hundred twenty-one completed questionnaires from passengers on three flights were collected. Of those who answered (n = 117), the age range was 20–81 years (median: 53, IQR: 36–64 years); 48 of 120 (40.0%) were male and 72 of 120 (60.0%) female. Just over half of respondents were British (n = 64/118; 54.2%), 25.4% (n = 30/118) were Chinese and 20.3% (n = 24/118) were 'Other'. Most respondents could read English fluently (n = 99/118; 84.0%), 14 were bilingual and four trilingual. Seventeen (14.4%) could only read Mandarin and 1 (1.0%) could only read Cantonese. None of the respondents had been to Wuhan city in mainland China in the 14 days before arriving at LHR.

Knowledge of symptoms and actions to take

Most respondents correctly identified a fever/high temperature (87.6%), difficulty breathing (87.6%) and cough (85.1%) as the symptoms associated with COVID-19 (Table 1). In line with the official case definition at the time (described in PHE leaflets as cough, fever or shortness of breath), 101 (83%) of 121 respondents identified all three symptoms as symptoms of COVID-19.

Most participants were correctly aware that people with COVID-19 might not show symptoms immediately (77.1%) and that asymptomatic status could last for 14 days (75.4%). Of all participants, 92.4% of participants also thought that people with COVID-19 can be contagious even without symptoms. A minority of respondents (9.3%) mistakenly thought antibiotics could treat COVID-19, and a substantial proportion (27.1%) were uncertain.

Table 2 shows that most passengers were able to identify the recommended actions to take if they had been to Wuhan in the previous 14 days – to self-isolate (96.6%) and call NHS 111 for advice (84.6%). Respondents were less confident about actions to take for those who had visited other named destinations; among people who had travelled to Singapore in the past 14 days, most correctly stated that they should not take any action if well, in accordance with PHE information, but a substantial minority thought they should self-isolate (23.7%) and call NHS 111 for advice (18.8%), respectively, whereas the PHE leaflets advised these actions only for those with symptoms.

Attitudes to official advice

One hundred four of 121 (86.0%) passengers stated that they had read the leaflet (94 read the English version, 30 read the Mandarin version and 20 read it in both languages). Only 6 (5.0%) stated that they had not read it in either language.

Overall, respondents thought the leaflet and poster (leaflets distributed in flight had the same content as leaflets and posters displayed at the airport) were easy to understand (84.4% agree or strongly agree) and trustworthy (84.2% agree or strongly agree). Most respondents also agreed that they had received sufficient information on what to do in response to COVID-19 symptoms, including how and when to avoid contact with others (Table 3).

Qualitative findings

Fifteen interviews were conducted; five men and 10 women with ages ranging from 21 years to older than 80 years. Six were retired, five worked full-time, three were full-time students and one was unemployed. Most participants were permanent residents in the UK; three were limited-duration residents and two were temporary visitors. Most (11 participants) were British, three were Chinese, and one was from New Zealand. All Chinese participants could speak Mandarin and English and had seen PHE information in both languages. All White participants could speak only English.

The results represent passengers' views and perspectives on the public health advice and their experiences of self-isolation. These views clustered into five broad themes (Table 4). Only themes

Table 1
Recognition of COVID-19 symptoms in a sample of 121 passengers arriving at London Heathrow airport from COVID-19 affected countries between 4th and 13th March, 2020.

Symptom	Yes N (%)	No N (%)
Symptoms listed in PHE information		
Fever/high temperature	106 (87.6)	14 (11.6)
Difficulty breathing	106 (87.6)	14 (11.6)
Cough	103 (85.1)	18 (14.9)
Symptoms not listed in the PHE information		
Fatigue or tiredness	70 (57.9)	48 (39.7)
Sore throat	64 (52.9)	51 (42.2)
Sneezing	60 (49.6)	56 (46.3)
Runny nose	55 (45.5)	62 (51.2)
Chills/shivering	54 (44.6)	59 (48.8)
Aches or pains in your muscles, joints or bones	53 (43.8)	61 (50.4)
Headache	50 (41.3)	65 (53.7)
Loss of appetite	38 (31.4)	73 (60.3)
Nausea/vomiting	32 (26.5)	83 (68.6)
Diarrhoea	30 (24.8)	86 (71.1)
Stomach ache	14 (11.6)	100 (82.6)

Note: Percentages in Table 1 treat 'missing' as another group since 'Not sure' was not an option offered for this question.

Table 2
Knowledge of health-seeking behaviour in a sample of 121 passengers arriving at London Heathrow airport from COVID-19–affected countries between 4th and 13th March, 2020.

Statement	True N (%)	False N (%)	Not sure N (%)
Statement advised in PHE information			
If someone arriving in the UK has been to Wuhan in mainland China in the past 14 days, they should stay indoors and avoid contact with others	114 (96.6)	3 (2.5)	1 (0.9)
If someone arriving in the UK has been to Wuhan in mainland China in the past 14 days, they should call NHS 111 for advice	99 (84.6)	12 (10.3)	6 (5.1)
Statement not advised in PHE information			
If someone arriving in the UK has been to Singapore in the past 14 days, they should stay indoors and avoid contact with others	28 (23.7)	75 (63.6)	15 (12.7)
If someone arriving in the UK has been to Singapore in the past 14 days, they should call NHS 111 for advice	22 (18.8)	80 (68.4)	15 (12.8)

Note: Percentages are for those who responded to the statement.

Table 3
Attitudes to official Public Health England advice in a sample of 104 passengers arriving at London Heathrow airport from COVID-19–affected countries between 4th and 13th March, 2020.

Statement	Strongly disagree N (%)	Mostly disagree N (%)	Mostly agree N (%)	Strongly agree N (%)
The leaflet and poster at the UK airport were easy to understand	10 (10.4)	5 (5.2)	36 (37.5)	45 (46.9)
The leaflet and poster at the UK airport can be trusted	11 (11.6)	4 (4.2)	36 (37.9)	44 (46.3)
I have received enough information about what to do if I develop symptoms of coronavirus	13 (12.2)	4 (3.7)	35 (32.7)	55 (51.4)
I have received enough information about how and when to avoid contact with other people	12 (11.1)	5 (4.6)	44 (40.7)	47 (43.5)

Note: Percentages are for those who responded to the statement.

relating directly to the reception of public health advice are reported below.

Knowledge of symptoms and actions to take if symptomatic

Thirteen of 15 participants recalled receiving the information leaflet during the flight or at the airport in Singapore or China. Most were impressed with the information and measures being taken at departure airports and surprised that '*there was almost nothing*' [participant 11] and '*nobody seemed to care*' [participant 8] on arrival at LHR. Only three passengers saw posters, which they said were not eye-catching (Table 5, quote 1; Fig. 1).

Cough, fever/high temperature and, progressively, breathing difficulties were the most frequently mentioned symptoms; '*you may be asymptomatic and so you have a cough or you might come down with a full-blown fever to the point where really you cannot breathe*' [participant 1]. Many passengers associated other diverse symptoms such as headache, fatigue, loss of smell and taste with COVID-19 although they were not included in the official case definition at the time.

Most participants said they would start with self-isolation when symptoms were mild and call NHS 111 if symptoms progress, indicating they would follow official advice and base their actions on disease severity (Table 5, quote 2).

Attitudes to official advice

The content of UK official advice was considered reasonable and adequate; passengers commented that it was '*quite clear and sensible*' [participant 5] and felt the government was taking some action in response to the outbreak.

Participants commonly mentioned concerns that people in the UK may disregard official advice, citing their lived experience in affected countries where televised public health information for COVID-19, including on social distancing and washing your hands, was '*reinforced every time there was a commercial break*', whereas in the UK '*it'is random*' [participant 2]. They noted that the lack of visible pandemic control measures at LHR gave '*a false sense of security*' [participant 7] and suggested reinforcing official measures such as installing temperature scanners, handing out materials and increasing number of personnel at airports, as well as enacting compulsory regulations to limit close contact and quarantine arrivals (Table 5, quotes 3 and 4).

Acting on official advice

Most participants had acquired information from both the UK and countries of departure, regardless of their usual country of residence. Since COVID-19 had already spread in the countries where travel originated, participants considered they were '*educated enough about it*' [participant 7] and treated it more seriously than the UK population; they were, as one participant put it, '*a bit ahead of the game*' [participant 3]. On arrival in the UK, as a precaution many participants voluntarily self-isolated or tried to distance themselves and avoided activities where people would be gathering, although this was not officially advised at that time (Table 5, quotes 5 and 6).

Table 4
Themes and subthemes related to passengers' views on public health advice and self-isolation.

Themes	Subthemes
Understandings related to COVID-19	COVID-19 knowledge/personal or lived experience/exposure/domestic concerns/personal protective equipment
Attitudes towards information materials and presence, self-isolation and lockdown	Attitudes on advice, information and presence/attitudes on self-isolation and lockdown/public adherence and perceptions of other/social pressure
Practices and experience during the pandemic	Difficulties/feeling lucky/self-disciplinary/compulsory measures
Information and advice	UK official advice/other source information/clear/reliability
Support	Emotional support/healthcare support/information support/instrumental support

Participants expressed awareness of their exposure risk while travelling that led some of them to self-isolate (see Table 5, quotes 7–9). They further noted that by doing so, they would avoid blame if any of their loved ones did get sick; one said they knew there was likely to be a 'stigma' around them having come from an affected country [participant 10].

Despite experiencing some mental pressure, participants expressed feeling fortunate to have the physical and social resources to manage their self-isolation effectively, while being aware that this was not the case for everyone (Table 5, quote 10).

The reasonable and clear official information was seen to shape public understanding of the COVID-19 crisis and therefore as promoting public acceptance of official advice (Table 5, quote 11). Participants further emphasised the crucial role of community support; '*I think providing they have sufficient support in their communities there is no reason at all why anybody should not self-isolate*' [participant 9].

Differences between Chinese and British passengers

Regarding advice about calling NHS 111, Chinese respondents shared more concerns than British respondents, including difficulties in getting through to an advisor, the vagueness of advice itself and uncertainty about whether NHS support is available for non-citizens. Alongside calling NHS 111, while some British respondents noted contacting their GP as a potential source of advice, Chinese respondents relied more on personal/social networks, such as teachers or supervisors (Table 5, quote 12). Chinese passengers further noted that, compared with China, people in the UK follow advice on an entirely voluntary basis. One Chinese respondent suggested that '*self-isolation must be compulsory*' [participant 15]; otherwise it will not be universally enacted by the public even if the advice itself is good.

Chinese passengers and British passengers have contradictory views on wearing face masks. Chinese respondents suggested to add wearing masks into UK official advice and despite their awareness of cultural and policy differences, emphasised their concerns that staff at the airport did not wear masks (Table 5, quote 13). Conversely, the majority British respondents noted their lack of conviction in the use of masks due to the absence of clear evidence (Table 5, quote 14). Some were actively opposed to the use of masks because '*they could do more harm than good*' [participant 3].

Discussion

Our findings show that passengers arriving from China and Singapore in the containment phase of the COVID-19 pandemic found the content of official public health information from PHE to be clear and easy to understand. Most correctly identified the actions to be taken when becoming symptomatic or arriving from certain destinations and considered this advice to be acceptable and trustworthy. However, there was some uncertainty regarding whether those arriving from a country or territory listed in PHE information other than Hubei or Wuhan should self-isolate or call the NHS helpline. Most of those surveyed (83%) correctly identified all three symptoms described in the leaflets and poster, but over half those surveyed and many of those interviewed also identified fatigue and sore throat as symptoms, with substantial proportions identifying other symptoms not included in the official case definition during the evaluation period. This definition changed over time alongside evolving scientific knowledge of the virus, and some symptoms identified by respondents have since been recognised as common manifestations of COVID-19, including anosmia which is now included in the official case definition. Because these passengers were arriving from countries where COVID-19 had spread

Table 5
Passengers' views and perspectives on public health advice (illustrative quotes).

Number	Quotes
1	*'I walked fast passing (those leaflets/posters), didn't pay much attention.'* [participant 15]
2	'*Well the first thing I would have had to have done would be to self-isolate. … And if the symptoms obviously got progressively worse I would then either contact my GP or phone 111. But it's a fairly straightforward process that's been set up to do this*'. [participant 9]
3	'*At Heathrow, we arrived and it was like nothing was wrong in the UK, so I think that causes a false sense of security, so maybe if there was more of a presence, like information, temperature check, personnel etc, people might take it more seriously.*' [participant 5]
4	'*Well they could have had thermal imaging cameras, they could have had medical staff in protective clothing there to talk to people whose temperature came up as above the norm, they could have then asked people in those conditions, you know, if they met those conditions to isolate them, you know.*' [participant 8]
5	' *… even though there wasn't the, you know, that wasn't really about the distancing over here, but we just thought we won't see family and friends for some time just because we'd been or gone through Singapore.*' [participant 3]
6	'*I didn't dare go to the university to take the exam on Monday, because the teacher said if you didn't feel well you could stay at home and didn't have to go to the university to take the exam.*' [participant 13]
7	' *… but being on the plane with other people coming from who knows where with who knows what, you know, we were a bit more concerned which is why we isolated when we came home.*' [participant 11]
8	'*We didn't want to put any of our family members or friends at risk in case we were carrying the virus but didn't know it.*' [participate 2]
9	' *… we sort of knew pretty much that the chances of us giving him (family member) anything were miniscule, because we wouldn't have put anybody at risk if we really thought that there was a chance but we just didn't want it on us'.* [participant 3]
10	'*I can't think about it, I have to think about we're very lucky, we're luckier than most and if I want to go down and walk along the beach I sort of can. … I think if somebody is locked up in a one-bedroom flat in London it will be horrible, it must be horrible for them …* ' [participant 10]
11	'*I can't think why you would not follow the official advice but I think the mere … at the time the number of people who had died from Coronavirus it was rising but I think … and the numbers were unclear, but they were talking about one to two percent of the people who got infected may die …* ' [Pprticipant 1]
12	'*Someone told me it [NHS 111] is constantly engaged … I would have hoped to know how to contact the NHS effectively in the case that I was infected. At that time, one could not get through to the NHS helpline. Maybe I could have been given a few more telephone numbers? This kind of information enabling me to have access to medical treatment would have given me a sense of security.*' [participant 15]
13	'*They [airport staff at customs] told us to take our face masks off. I understood their request. But the staff there didn't wear face masks. … As far as face masks are concerned, it is said that perhaps the virus will spread faster when face masks are not worn.*' [participant 14]
14	' *… and there wasn't any clear evidence to say a mask, an ordinary mask would prevent you picking up germs and if you did pick up a germ it would multiply inside the mask. So even though we had masks in our bags … so we had everything with us but we decided we'd use the hand gel but we didn't want to wear the masks.*' [participant 8]

further than in England when the study was conducted,[9,10] their responses may well reflect knowledge acquired elsewhere.

Support for this is shown by the fact that while most survey respondents indicated they had received sufficient information both about what to do if symptoms developed and about how and when to avoid contact with other people, the PHE leaflets and posters provided no information on avoidance of contact, beyond the requirement to stay indoors if symptomatic or when arriving from specified source locations. Our interview data support the survey findings that respondents believed the official information was adequate; however, their accounts show that respondents' knowledge was substantially informed by familiarity with public health interventions being taken elsewhere to contain transmission. For these passengers, the lack of visible infection control measures on arrival into the UK indicated a worrying lack of official concern about COVID-19. Their comments were verified by our researchers' observations that the design and positioning of PHE information at the arrival airport made it largely unnoticeable to arriving passengers (see Fig. 1), and by other studies highlighting a rejection of 'eye-catching measures' in the UK at the beginning of the outbreak.[11] Passengers' expressions of concern indicate that although the intended purpose of the leaflets was to provide information and guidance that would encourage people to follow recommended behaviours, recipients saw information provision along with other observable public health measures as an index of the adequacy of governmental outbreak response. The advice and information we evaluated thus served two roles – its intended function of public health messaging, and a reflection of the performance of official authorities. When passengers are already well informed by prior acquisition of knowledge elsewhere, as in our sample, they seem more concerned with its role as indicative of public health performance.

pandemic. Our respondents highlighted their own self-discipline not only in following official advice to self-isolate when advised but also in some cases going beyond it by self-isolating as a self-initiated precautionary measure. This action was linked to perceptions of exposure risk in affected countries where travel originated or during the journey and to concerns about stigmatisation should family or colleagues subsequently become infected. Similar findings have been reported by previous studies[13,14] indicating that travellers arriving from Ebola-affected countries restricted movement to avoid community stigmatisation. The additional interventions advocated by our respondents and their reported behaviours suggest screening people at entry, as done in 'enhanced screening' for Ebola, may help to reassure the travelling public that containment measures are in place. One recent study showed that, compared with no control, screening at entry, particularly through testing and isolating test-positive cases, can significantly reduce COVID-19 case importation numbers.[15] However, these screening measures generate other difficulties such as availability of testing kits and staff,[15] the length of time required to receive test results, how to maintain high sensitivity and accuracy,[15] and how to accurately target passengers and avoid social stigma.[6,16] Although quarantine for all arrivals could be another useful way to prevent the entry of infection if effective testing practices are not established, its efficacy will be affected by the length and location of quarantine, and longer duration quarantine entails a heavy burden even for resource-rich countries.[15,17] Currently there is significant cross-national variation in the use and enforcement of testing and quarantine measures alongside public health advice at border entry, creating widespread inconsistencies and potential confusion for travellers. The UK government currently requires passengers to (voluntarily) self-isolate at home for 10 days if arriving from an affected country but this can be ended earlier if a negative COVID-19 test result is obtained.[18]

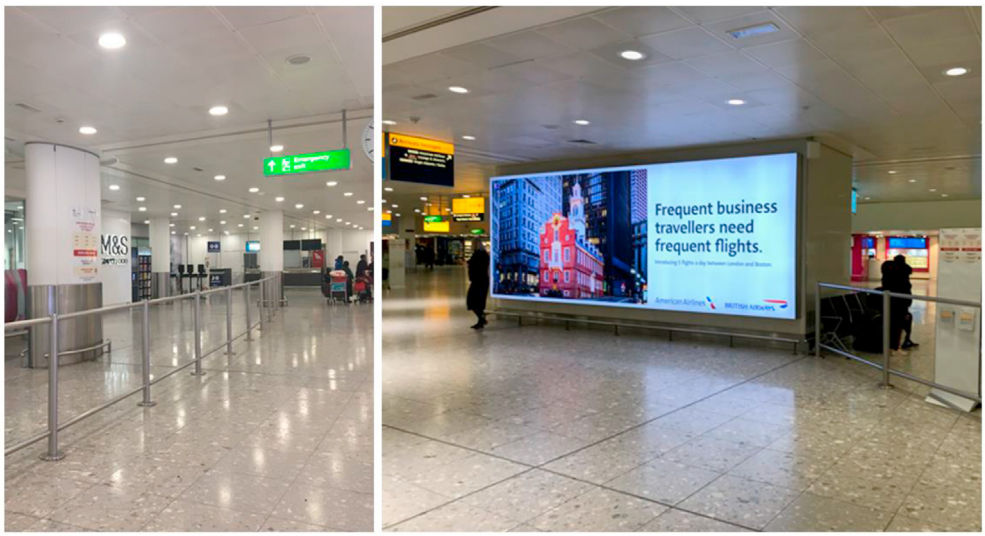

Fig. 1. Public Health England poster and leaflets, providing information and advice on COVID-19, at Terminal 3 arrivals, London Heathrow Airport in west London, 4 March 2020. Source: photos taken by researchers. Note: left, COVID-19 poster on pillar; right, COVID-19 leaflet stand (right hand side).

Our interviewees suggested various additional non-pharmaceutical interventions that were not in place on arrival, such as restricted contact tracing, temperature checks, widespread testing, and self-isolation/quarantine for all arrivals, many of which were then being used in countries such as Singapore and China and were eventually implemented in the UK.[12] This again indicates that passengers were using prior experience of pandemic control measures elsewhere to judge how seriously UK authorities were treating the

Respondents' good understanding of the information content of the PHE leaflet, which they received in flight, contrasted starkly with their reports of low visibility of, and minimal interaction with, similar materials on arrival. This suggests that providing public health information in flight, by announcements and distribution of written material – when passengers have the time to absorb it with few distractions – may be the more effective strategy. Chinese respondents commenting on PHE advice suggested that provision of

additional information and advice through departure countries or drawing on international perspectives could reassure non-citizen travellers who are not familiar with the UK healthcare system. The expressed concern of these respondents regarding mask use is vindicated by accumulating evidence and consequent changes in European policy. A recent review found a correlation between COVID-19 transmission events in flight and non-enforcement of rigid masking policy.[4] The UK government has mandated the use of face coverings in airports and on board commercial flights since the lifting of air travel restrictions in June 2020.[19] Further research is required to inform the evaluation of other potentially important strategies that could help to control infection risk and ease travel restrictions in the era of COVID-19, such as pretravel consultations that assess passengers' individual risk level and evaluate trip determinants in relation to COVID-19 policies in both origin and destination countries;[20] and the benefits, risks and acceptability of immunity passports that certify passengers as protected against COVID-19.[21,22]

This study has several limitations. Because this study was conducted in the early stages of the COVID-19 outbreak, owing to the geographical focus of the outbreak at that time rapid reductions in flights, our research was limited to a small number of flights from Asia. Broader representation of respondents from different nationalities with more geographic diversity of settings since the pandemic has progressed is needed in future studies. Study size and opportunities to use our findings to inform the content and delivery of official public health guidance were limited by difficulties gaining airside airport access and obtaining cooperation from airlines, so that by the time we implemented data collection, the number of passengers arriving from affected countries had diminished drastically. Interviewees' views might have changed between survey completion on arrival and interview due to time elapsed and rapid changes in pandemic and UK policies; all interviews were completed within seven weeks from arrival date to minimise these effects. Finally, our respondents' observations regarding public health advice on arrival into the UK are inherently time-limited, in view of the rapidly changing pandemic and associated public health policy. Nonetheless, six months after the completion of our data collection, following the resumption of international travel to and from the UK, international travellers were still reporting a lack of visible public health measures or active enforcement of self-isolation regulations on arrival.

Conclusion

Our findings confirm the clarity and acceptability of public health guidance on COVID-19 provided to passengers arriving into UK ports in the early stages of the pandemic. They also demonstrate a widespread perception that information provision alone was an insufficient official response to this global public health emergency. This is cause for concern since it may reduce trust in official sources, an established driver of non-adherence to public health interventions.[23] It also indicates that public health information provision at borders should be appraised not only for its functional effectiveness in imparting guidance and encouraging behaviours to control transmission, but also for its perceived effectiveness in furnishing public assurance of official action to contain the disease threat. Travellers arriving from countries where COVID-19 was already established frequently had knowledge of the disease and of transmission containment measures not derived from official UK advice or present in the UK at that stage. In a rapidly evolving international health crisis, particularly one in which understanding of the disease is partial and changing, evaluating public understanding by reference to locally defined parameters can be unreliable, especially as knowledge among those with experience from elsewhere may be more advanced than local understanding. This indicates the value of appraising public perceptions not only to measure understanding and adherence but also to gain insights into future potential measures and their likely acceptability. Our study also demonstrates the complexity of health policy decision-making in international public health emergencies and provides fresh insights into the need to take account of the diverse information sources on which international travellers may draw. Finally, it highlights the importance of establishing more efficient mechanisms for rapid appraisal and feedback to public health and regulatory authorities of social science evidence that could contribute to containment and control of epidemic disease threats.

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Ethical approval

This study was a form of service evaluation and PHE's ethics committee, the PHE Research Ethics and Governance Group, confirmed that no ethical approval was required.

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Competing interests

None declared.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.puhe.2021.01.028.

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