Inclusivity in Global Health Research Priority Setting: Can we do better? Lessons from CHNRI Cleft

Niki Kouvroukoglou, Sanita Sandhu, Barbara Delage, Debbie Sell, Nicola Stock, Gareth Davies, Marina Campodocino, Bruce Richard, Zipporah Njeri Gathuya, Mekonen Eshete, David Fitzimons, Orla Duncan, Felicity Vidya Mehendale

Email: s1807091@ed.ac.uk

THE UNIVERSITY of EDINBURGH | usher institute

Introduction

Why conduct research prioritisation exercises?

In 1990 the CHRD report highlighted that only 5% of available global health research resources were invested in Low-and-Middle Income Countries (LMIC) where 93% of preventable deaths occurred. [1]
As a result many tools for research prioritisation were developed.

A methodological review in 2016 found that the most popular tool was the Child Health and Nutrition Research Initiative (CHNRI) method (26%) [3].

What is the CHNRI method?

The CHNRI method was developed in 2007 as a transparent framework to conduct research priority-setting exercises. [4] It is built upon crowdsourcing. where experts are invited to submit research questions they would like answered in the future.
These questions are then condensed and 200 of them are scored and ranked using certain criteria. Stakeholders can use this list of questions for their research interest and funding.

Why did we choose the CHNRI method?

Democratic and inclusive method [12]	Crowdsourcing allowing participation from a wide range of individuals
Transparency of the method [12]	Criteria set from the beginning of the scoring process
Global audience [12]	Multi-lingual and multidisciplinary approaches were necessary for our project and the CHNRI method had the capacity to do this.
Flexibility towards inclusivity	We needed to be inclusive to non-research focused clinicians/professionals, parents/patients and NGO representatives, This was not part of the CHNRI methodology, but it had the **flexibility** to allow for these changes.

Why conduct a research priority setting exercise in orofacial clefts?

To date, there are no published CHNRI exercises in orofacial clefts (OFC). OFCs are the most common congenital condition affecting the face and head with an incidence of approximately 1 in 700 live births worldwide, with significant global variation in the burden of disease, management, and outcomes [6]. Other priority-setting exercises exist but none on a global scale.

Previous priority setting exercises in orofacial clefts include:

UK-wide James Lind alliance focus-group approach [7]
Workshop in 2007 on future research priorities for OFCs [8]
Social-media-based research prioritisation for multiple congenital abnormalities [9]
Prioritasation on sleep-disordered breathing in OFCs [10]
A Delphi study on the management of otitis media with effusion in cleft palate patients [11]

Aims:

- To describe changes of the CHNRI methodology carried out to enhance inclusivity of research priority setting exercises on a global scale.
- To report the outcome of this approach and ongoing challenges.

Methods

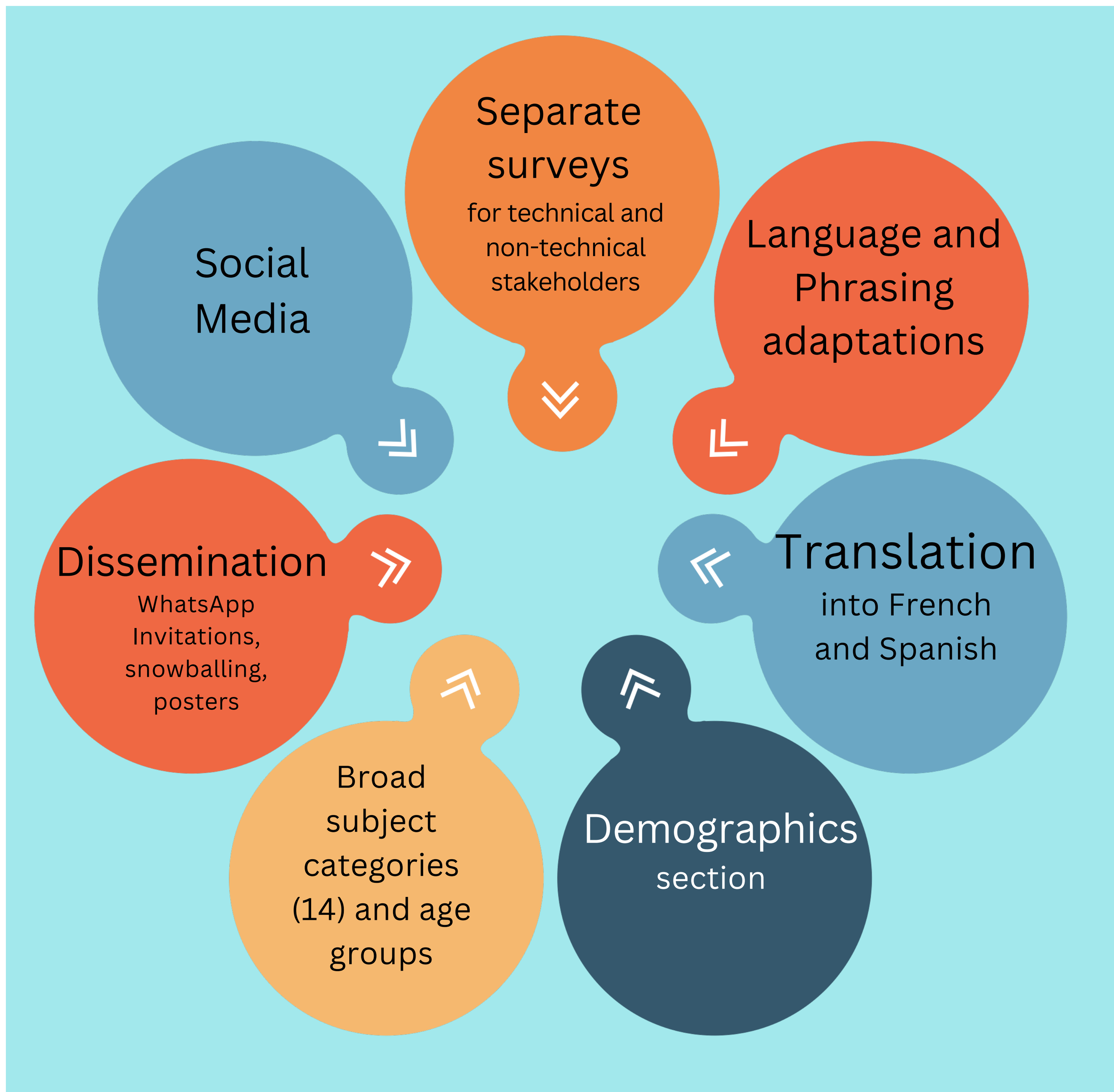
Data management and Ethics

The survey was created using the online Joint Information Systems Committee (JISC) platform.
It allowed for multiple collaborators, easy editing, easily downloadable data and was also secure.

The Steering Group

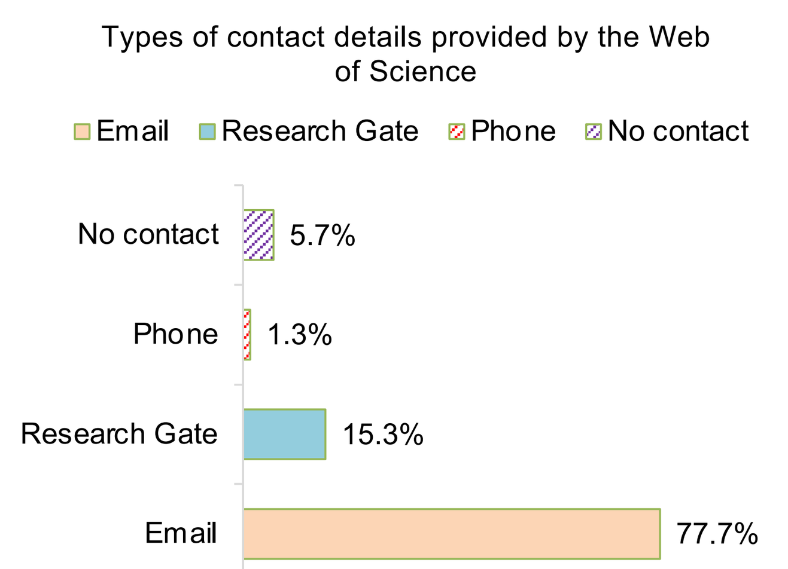
This exercise gathered a group of 15 technical experts with interest in OFC research. Individuals from a mix of backgrounds were invited (surgeons, dentists, nutritionists, program leaders) and from multiple countries (both high and LMICs) to incorporate a diversity of views from the wider research community.
Their role was to provide input on how to increase inclusivity in this CHNRI exercise

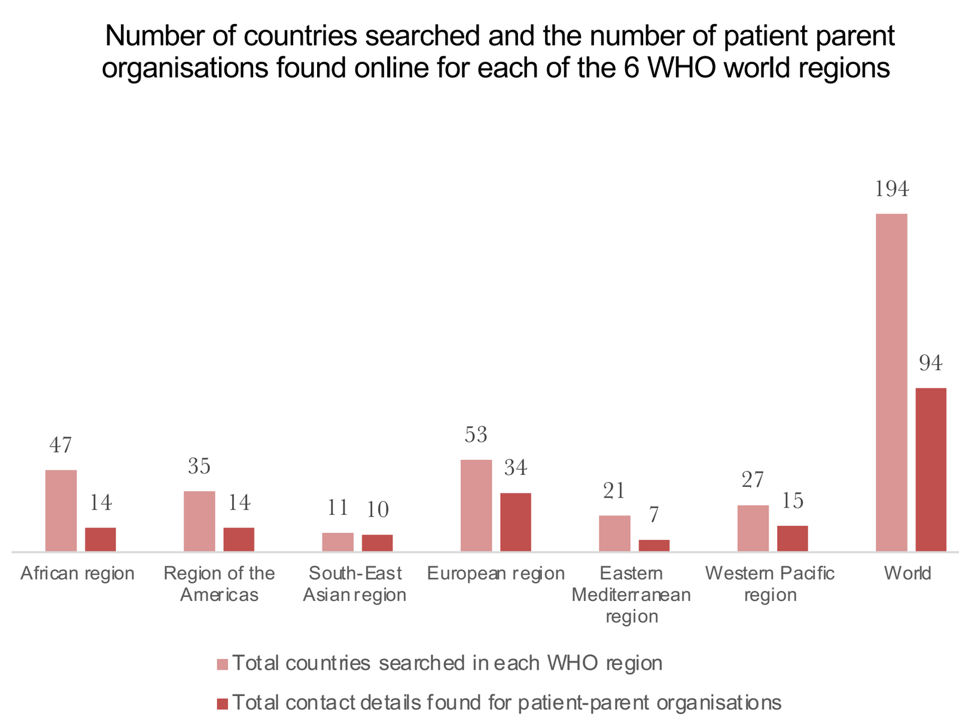
Modifications to improve inclusivity



Collaborator recruitment

Web of Science search provided surnames and initials of the **300 most published researchers in the field of cleft lip and palate.**
Google and Bing searches were performed to find contact information for these researchers.
Contact information included email addresses, Research Gate accounts and phone numbers. 233 email addresses and 46 Research Gate accounts were collected.
National NGO and patient parent organisations contact information was also researched for every country in the world. In total 94 organisations were found and contacted.





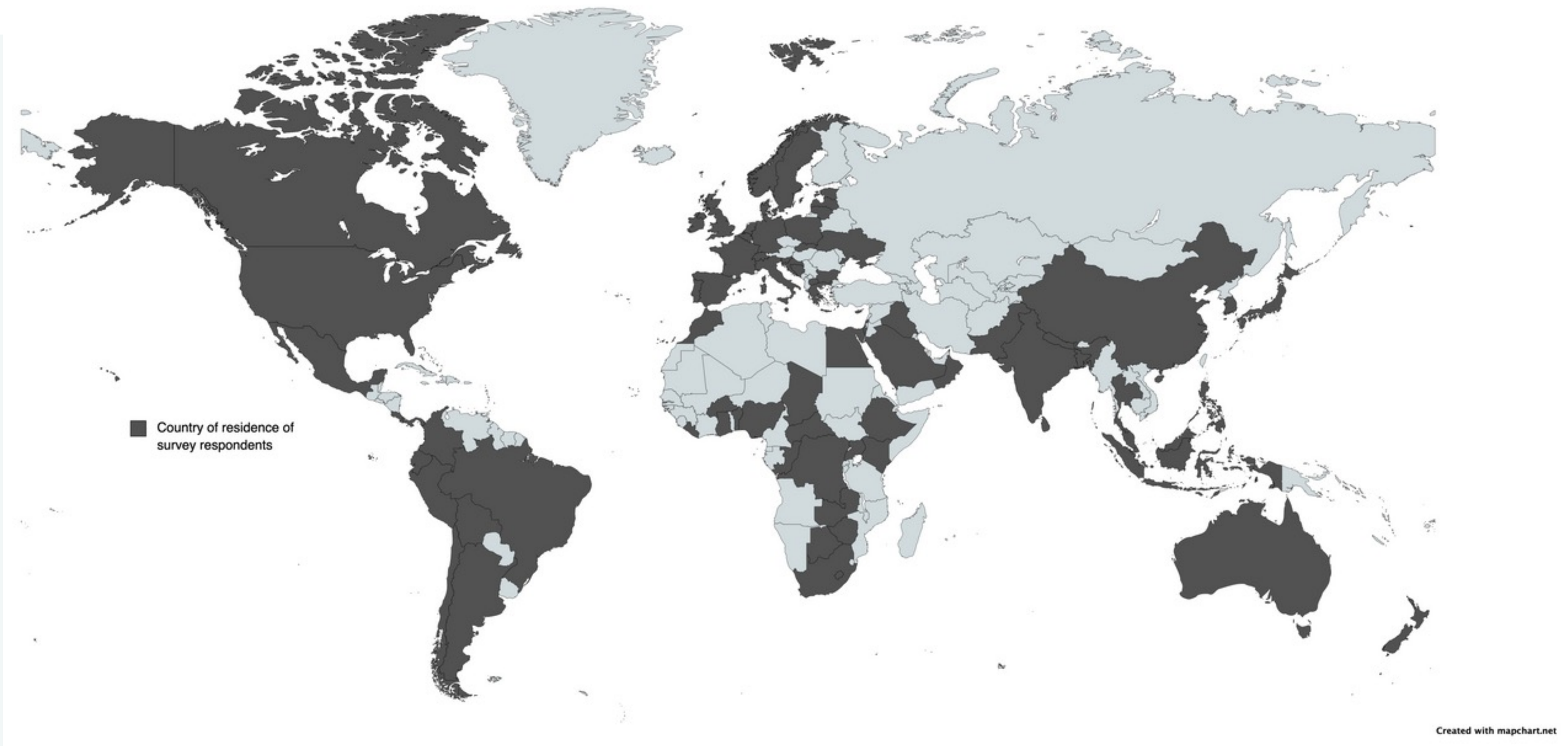
Results

Overall Numbers and Global reach

412 collaborators answered the survey
1422 questions were submitted

The survey was open over a period of five and a half months

78 countries were reached comprising 40% of the countries in the world.



Gender

56% of respondents were female and 44% were male.

Specialty

Top specialties
Surgeons comprised the largest group of collaborators with a 30.42% share. This was followed by orthodontists (10.21%) and speech therapists (8.54%).
Underrepresented specialties:
Collaborators belonging to the medical specialties of anaesthetists, paediatricians and general practice were under-represented in the survey. They took 3.33%, 2.50% and 0.21% of the vote respectively.
Non-technical stakeholders:
Members of patient parent organisations/non-governmental organisations comprised 8.96% of the overall share and family members of individuals with OFCs comprised 6.67%.

Broad categories

The largest group was questions about surgery which made up **12%**
There were no groups significantly more under-represented than another.
The "Other" category made up 5% of overall questions and they included: 'aetiology of clefts', 'resource management', 'all', 'prenatal care counselling' and 'quality of life'.

Discussion

Previous CHNRI exercises have invited experts, some including clinicians, to submit questions but this is one of the first CHNRI exercises to invite stakeholders from non-technical backgrounds and national parent organisations and NGOs to propose research ideas. [13,18-21]

This CHNRI exercise has more responses than any other global CHNRI exercise.

Online dissemination of surveys using social media has been successful in the past and was also used in our methodology.

Language translation in 3 languages. This could have led to limitations as we did not include some common native languages (Mandarin, Hindu, Arabic etc.). [24]

Use of WhatsApp as a method of dissemination was deemed "intrusive and non-conventional" by the Ethics committee even if it was the preferred method of contact for many busy clinicians in LMICs.

9% of the submitted questions were not phrased in a traditional research question format, i.e., a coherent sentence followed by a question mark. This could be the result of promoting inclusivity above all and gathering responses from non-research active individuals

Patient parent organisations without online activity could not be contacted.

We anticipated that surgeons were likely to be the largest group of respondents and adopted approaches to improve representation of other disciplines. Despite this, it remains challenging to involve groups that are non-surgical and non-clinical and it has highlighted how important this effort is.

References

1. Evans J, C.G., Abed F, Bergstrom S, Calloway D, Ezzat E, et. al., Health Research: Essential link to equity in Development., C.o.h.r.f.d. [Internet]. Editor. 1990, Commission on Health Rsearch for Development: Oxford.
2. Development, C.C.o.H.R.f., Essential National Health Research and Priority Setting: Lessons Learned. 1997, Council on Health Research for Development: [Internet].
3. Yoshida, S., Approaches, tools and methods used for setting priorities in health research in the 21(st) century. J Glob Health, 2016. 6(1): p. 010507.
4. Rudan, I., et al., Setting health research priorities using the CHNRI method: VII. A review of the first 50 applications of the CHNRI method. J Glob Health, 2017. 7(1): p. 011004.
5. Rudan, I., et al., Setting priorities in global child health research investments: guidelines for implementation of CHNRI method. Croat Med J, 2008. 49(6): p. 720-33.
6. Agbenorku, P., Orofacial Clefts: A Worldwide Review of the Problem. ISRN Plastic Surgery, 2013. 2013: p. 7 pages.
7. Petit-Zeman, S. and K. Cowan, Patients/carers and clinicians can set joint priorities for research in cleft lip and palate. International Journal of Pediatric Otorhinolaryngology, 2013. 77(3): p. 309-310.
8. Yazdy, M.M., et al., Priorities for Future Public Health Research in Orofacial Clefts. The Cleft Palate Craniofacial Journal, 2007. 44(4): p. 351-357.
9. Sinclair, M., et al., Exploring Research Priorities of Parents Who Have Children With Down Syndrome, Cleft Lip With or Without Cleft Palate, Congenital Heart Defects, and Spina Bifida Using ConnectEpeople: A Social Media Coproduction Research Study. Journal of Medical Internet Research, 2019. 21(11).
10. Davies, K., et al., Parental Experience of Sleep-Disordered Breathing in Infants With Cleft Palate: Comparing Parental and Clinical Priorities. Cleft Palate-Craniofacial Journal, 2019. 56(2): p. 222-230.
11. Harman, N.L., et al., MOMENT – Management of Otitis Media with Effusion in Cleft Palate: protocol for a systematic review of the literature and identification of a core outcome set using a Delphi survey. Trials, 2013. 14(1): p. 70.
12. Rudan, I., Setting health research priorities using the CHNRI method: IV. Key conceptual advances. J Glob Health, 2016. 6(1): p. 010501.
13. Shah, H., et al., Research priorities to reduce the global burden of dementia by 2025. The Lancet Neurology, 2016. 15(12): p. 1285-1294.
14. Yoshida, S., et al., Setting health research priorities using the CHNRI method: II. Involving researchers. J Glob Health, 2016. 6(1): p. 010302.
15. Kapiriri, L., et al., Setting priorities in global child health research investments: addressing values of stakeholders. Croat Med J, 2007. 48(5): p. 618-27.
16. Sandhu, S.K., et al., Lost in translation: a caution against using automated and commercial translation services for medical communications. BMJ Global Health, 2022. 7(1): p. e008067.
17. Williams, K., et al., Standard 6: age groups for pediatric trials. Pediatrics, 2012. 129 Suppl 3: p. S153-60.
18. Korte, M., et al., Setting a research agenda to advance maternal, newborn, and child health in Ethiopia: An adapted CHNRI prioritization exercise. Journal of Global Health, 2023. 13.
19. Paskins, Z., et al., Research priorities to reduce the impact of musculoskeletal disorders: a priority setting exercise with the child health and nutrition research initiative method. Lancet Rheumatol, 2022. 4(9): p. e635-e645.
20. Chapman, N., et al., Setting national research priorities for difficult-to-treat depression in the UK between 2021-2026. J Glob Health, 2022. 12: p. 09004.
21. Adeloye, D., et al., Research priorities to address the global burden of chronic obstructive pulmonary disease (COPD) in the next decade. J Glob Health, 2021. 11: p. 15003.
22. Krist, A.H., et al., Engaging Patients in Decision-Making and Behavior Change to Promote Prevention. Stud Health Technol Inform, 2017. 240: p. 284-302.
23. Bombard, Y., et al., Engaging patients to improve quality of care: a systematic review. Implement Sci, 2018. 13(1): p. 98.
24. Irvine, C., et al., Setting Global Research Priorities in Pediatric and Adolescent HIV Using the Child Health and Nutrition Research Initiative (CHNRI) Methodology. J Acquir Immune Defic Syndr, 2018. 78 Suppl 1(1): p. S3-s9.
25. Sivaratnam, S., et al., Using Social Media to Engage Knowledge Users in Health Research Priority Setting: Scoping Review. J Med Internet Res, 2022. 24(2): p. e29821.
26. Tomlinson, M., et al., Global research priorities to accelerate programming to improve early childhood development in the sustainable development era: a CHNRI exercise. Journal of global health, 2019. 9(3): p. 020703-020703.
27. De Man, J., et al., The pandemic of online research in times of COVID-19. BMJ Open, 2021. 11(2): p. e043866.
28. de Koning, R., et al., Survey Fatigue During the COVID-19 Pandemic: An Analysis of Neurosurgery Survey Response Rates. Front Surg, 2021. 8: p. 690680.