

University of Groningen

Global investments to optimise the health and wellbeing of children with disabilities

Olusanya, Bolajoko O.; Davis, Adrian C.; Hadders-Algra, Mijna; Wright, Scott M.

Published in:
The Lancet

DOI:
[10.1016/S0140-6736\(22\)02368-6](https://doi.org/10.1016/S0140-6736(22)02368-6)

IMPORTANT NOTE: You are advised to consult the publisher's version (publisher's PDF) if you wish to cite from it. Please check the document version below.

Document Version
Publisher's PDF, also known as Version of record

Publication date:
2023

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):

Olusanya, B. O., Davis, A. C., Hadders-Algra, M., & Wright, S. M. (2023). Global investments to optimise the health and wellbeing of children with disabilities: a call to action. *The Lancet*, 401(10372), 175-177. [https://doi.org/10.1016/S0140-6736\(22\)02368-6](https://doi.org/10.1016/S0140-6736(22)02368-6)

Copyright

Other than for strictly personal use, it is not permitted to download or to forward/distribute the text or part of it without the consent of the author(s) and/or copyright holder(s), unless the work is under an open content license (like Creative Commons).

The publication may also be distributed here under the terms of Article 25fa of the Dutch Copyright Act, indicated by the "Taverne" license. More information can be found on the University of Groningen website: <https://www.rug.nl/library/open-access/self-archiving-pure/taverne-amendment>.

Take-down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Downloaded from the University of Groningen/UMCG research database (Pure): <http://www.rug.nl/research/portal>. For technical reasons the number of authors shown on this cover page is limited to 10 maximum.

Moving forward, we here offer three general suggestions for producing machine learning markers with maximal impact. First, focus should be cast on modifiable risk. For example, an algorithm that uniformly assigns high risk of myocardial infarction to older individuals is likely to be less valuable than one which accurately predicts risk in a 30-year-old, where risk factor modification has greater potential to alter disease trajectory. Second, biologically aware data modalities should be leveraged. Imaging and omics features, particularly when data are acquired more uniformly (eg, electrocardiograms, echocardiograms, genomic arrays) might be less subject to ascertainment biases compared with other types of EHR variables. Third, gaps need to be filled in with regard to existing standards of care. An important consideration, particularly for complex models, is whether increased predictive performance provides an adequate improvement over the many risk models already available. This is particularly true for coronary artery disease, for which previous studies have noted comparable risk performance using laboratory-based and non-laboratory-based risk assessment,¹¹ stratification available from the time of birth based on a polygenic score to quantify inherited susceptibility,¹² and considerable utility of a non-invasive CT scan to measure coronary artery calcification.¹³

With the growing availability of large datasets and maturation of novel machine learning marker methods like those presented in Forrest and colleagues' Article, we anticipate an important shift from the development of methods to maximise predictive accuracy to those that enable equitable identification of individuals who might respond best to a therapeutic intervention.

AVK reports grants from IBM Research, during the conduct of the submitted work to research the topic of machine learning in cardiovascular disease. AVK also reports personal fees from, employment by, and equity in Verve Therapeutics, and personal fees from Amgen, Novartis, Silence Therapeutics, Korro Bio,

Foresite Labs, Third Rock Ventures, Color Health, Veritas International, Sarepta Therapeutics, and Ambry, outside of the submitted work. PB reports grants from IBM Research, grants from Bayer, personal fees from Prometheus Bio, Flagship Pioneering, Recursion and Novartis, outside of the submitted work.

*Puneet Batra, Amit V Khera
pbatra@broadinstitute.org

Machine Learning for Health Group (PB, AVK) and Cardiovascular Disease Initiative (PB), The Broad Institute of MIT & Harvard, Cambridge, MA 02139, USA; Verve Therapeutics, Boston, MA, USA (AVK); Division of Cardiology, Department of Medicine, Brigham and Women's Hospital, Boston, MA USA (AVK)

- 1 Attia ZI, Noseworthy PA, Lopez-Jimenez F, et al. An artificial intelligence-enabled ECG algorithm for the identification of patients with atrial fibrillation during sinus rhythm: a retrospective analysis of outcome prediction. *Lancet* 2019; **394**: 861–67.
- 2 Pirruccello JP, Chaffin MD, Chou EL, et al. Deep learning enables genetic analysis of the human thoracic aorta. *Nat Genet* 2022; **54**: 40–51.
- 3 Ulloa-Cerna AE, Jing L, Pfeifer JM, et al. rECHOmmend: an ECG-based machine learning approach for identifying patients at increased risk of undiagnosed structural heart disease detectable by echocardiography. *Circulation* 2022; **146**: 36–47.
- 4 Goff DC Jr, Lloyd-Jones DM, Bennett G, et al. 2013 ACC/AHA guideline on the assessment of cardiovascular risk: a report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines. *J Am Coll Cardiol* 2014; **63**: 2935–59.
- 5 Ridker PM, Cook NR. Statins: new American guidelines for prevention of cardiovascular disease. *Lancet* 2013; **382**: 1762–65.
- 6 Patel AP, Wang M, Kartoun U, Ng K, Khera AV. Quantifying and understanding the higher risk of atherosclerotic cardiovascular disease among south Asian individuals: results from the UK Biobank Prospective Cohort Study. *Circulation* 2021; **144**: 410–22.
- 7 Forrest IS, Petrazzini BO, Duffy A, et al. Machine learning-based marker for coronary artery disease: derivation and validation in two longitudinal cohorts. *Lancet* 2022; published online Dec 20. [https://doi.org/10.1016/S0140-6736\(22\)02079-7](https://doi.org/10.1016/S0140-6736(22)02079-7).
- 8 Petrazzini BO, Chaudhary K, Márquez-Luna C, et al. Coronary risk estimation based on clinical data in electronic health records. *J Am Coll Cardiol* 2022; **79**: 1155–66.
- 9 Beaulieu-Jones BK, Yuan W, Brat GA, et al. Machine learning for patient risk stratification: standing on, or looking over, the shoulders of clinicians? *NPJ Digit Med* 2021; **4**: 62.
- 10 Agrawal S, Klarqvist MDR, Emdin C, et al. Selection of 51 predictors from 13 782 candidate multimodal features using machine learning improves coronary artery disease prediction. *Patterns (N Y)* 2021; **2**: 100364.
- 11 Gaziano TA, Young CR, Fitzmaurice G, Atwood S, Gaziano JM. Laboratory-based versus non-laboratory-based method for assessment of cardiovascular disease risk: the NHANES I Follow-up Study cohort. *Lancet* 2008; **371**: 923–31.
- 12 Khera AV, Chaffin M, Aragam KG, et al. Genome-wide polygenic scores for common diseases identify individuals with risk equivalent to monogenic mutations. *Nat Genet* 2018; **50**: 1219–24.
- 13 Detrano R, Guerci AD, Carr JJ, et al. Coronary calcium as a predictor of coronary events in four racial or ethnic groups. *N Engl J Med* 2008; **358**: 1336–45.

Global investments to optimise the health and wellbeing of children with disabilities: a call to action



On Nov 20, 2022, we celebrate World Children's Day and the theme this year is inclusion, for every child. However, children with disabilities have received little attention from global health and development stakeholders.

Since the launch of the first comprehensive global health agenda in 2000 under the Millennium

Development Goals, the dearth of population data had hampered global policy, investment, and interventions for children with disabilities.¹ For policy makers, no data mean no problem, which translates into no action. As part of the concerted efforts by stakeholders to address this gap within the framework of the UN's Sustainable

Published Online
November 18, 2022
[https://doi.org/10.1016/S0140-6736\(22\)02368-6](https://doi.org/10.1016/S0140-6736(22)02368-6)



Phymant Studio/Getty Images

Development Goals (SDGs) 2015–30, UNICEF, in 2021, published a landmark report on the state of the world’s children with disabilities.² The report was intended to provide authoritative data on the global prevalence of disabilities in children aged 17 years or younger and to draw attention to the living experiences of these invisible children and their families. Estimates based on data from household surveys of parent-reported functional difficulties indicate that almost 240 million (one in ten) children and adolescents have moderate-to-severe disabilities globally, including 29 million children aged 0–4 years.² Estimates from the Global Burden of Disease Study suggest that around 50 million (one in 12) children younger than 5 years have mild-to-severe disabilities requiring some form of intervention.¹³

The UN Convention on the Rights of the Child (1989) recognises the rights of a child with disability to a full and decent life of dignity, self-reliance, and active participation in the society for the fullest possible social integration. However, childhood disability is still neglected, stigmatised, and associated with discrimination and isolation, and places considerable emotional and financial burden on families. The absence of services for early detection and appropriate intervention including financial support in many communities, particularly in low-income and middle-income countries (LMICs),²⁴ often invokes a sense of helplessness and hopelessness for parents about the uncertain future that awaits their child. According to UNICEF, children with disabilities have worse outcomes than children without disabilities in key measures of

early childhood care, health, and development as well as educational attainment.² For example, children with disabilities are 42% less likely to have foundational reading and numeracy skills, 49% more likely to have never attended school, 47% more likely to be out of primary school (children aged 6–11 years), 41% more likely to experience discrimination, and 20% less likely to have expectations of a better life.² These data suggest an urgent need for a robust global response to address the many avoidable health, educational, and social inequalities faced by children with disabilities and their families.

Investments to build local capacity in national health and educational systems to deliver early detection and intervention services for children with disabilities from early childhood offer pathways to wellbeing, optimal developmental outcomes, inclusive education, economic empowerment, financial independence, and the opportunity to become productive members of society.⁵ The provisions of the SDGs (4.2) for school readiness in early childhood towards equitable, inclusive, and quality education already present unequivocal political support to justify such investments as a priority for all countries. Development Assistance for Health (DAH; financial and in-kind contributions) is the major source of funding in global child health.⁶ Since 2015, an average of US\$8 billion annually has been expended on neonatal and child health services in LMICs representing about 20% of total DAH (\$40 billion) in 2019 before the COVID-19 pandemic. Understandably, these investments have so far been devoted entirely to strengthening the capacity of health systems to meet global targets for reducing child mortality. However, despite these substantial investments, health systems in many LMICs are still ill-equipped to support the needs of children with disabilities due to the scarcity of requisite facilities and qualified health-care professionals to deliver services.⁷ In April, 2022, the World Bank Group (WBG) launched the Childcare Incentive Fund in partnership with the governments of Australia, Canada, and the USA as well as some private donors to promote child development in LMICs.⁸ The US Agency for International Development has committed up to \$50 million over 5 years as part of the expected funding pool of at least \$180 million. No specific budget was announced for childhood disability. Even at the current level of funding for the care of children (which is less than 3% of annual expenditure on child survival) little or no progress will be achieved by 2030 for children with disabilities.

The available evidence on childhood disabilities calls for a recalibration of global funding priorities for a disability-inclusive child health agenda beyond survival.¹ A well focused global initiative to promote school readiness for disability-inclusive education as enshrined in the SDGs now needs to be accelerated as previously advocated.¹ School readiness requires investment to ensure the readiness of the individual child for primary school enrolment and participation; the school's readiness to provide an optimal learning environment for the child; and family and community support that contributes to child readiness for school.⁹ This process requires coordination between the health and educational sectors to ensure effective transition into the educational system at school entry.⁵ As a priority, sociocultural and financial access barriers to early detection and intervention must be addressed systematically to ensure that children with or at risk of disabilities are placed on a trajectory for optimum health, education, and development.^{2,10} Stigma and discrimination towards children with disabilities and their families are major sociocultural barriers that must receive attention.^{2,4,11} Intervention studies on stigma reduction reported so far are generally of low quality and call for further robust implementation research.¹¹ A 2022 *Lancet* Commission has offered a comprehensive framework for tackling stigma and discrimination in mental health that can be adapted effectively for childhood disability.¹² Investment in recruiting, training, and appropriately equipping health workers to routinely deliver basic screening services for conditions that can be readily detected from birth is also needed as part of health-care strengthening. For instance, a global programme to support early detection of children with hearing impairment and major birth defects in the first month of life as currently offered routinely in high-income countries should be considered as a starting point. The cost-effectiveness of neonatal hearing screening as a component of universal health coverage has been demonstrated.¹³ This global programme will entail investments to support the development of affordable assistive technologies including screening devices for LMICs.

The societal costs of not responding to these challenges and the population benefits of inclusion are substantial globally.^{14,15} Without purposeful and well coordinated global investments, many children will remain at risk of exclusion from inclusive education as envisioned by the SDGs. We are calling on the contributors to DAH, including

the WBG, to adjust their funding budget and priorities to reflect the stark inequity and inequality faced by children with disabilities in LMICs. The sooner the better.

All authors have lived experience of disability. All authors report being members of the Global Research on Developmental Disabilities Collaborators. MH-A reports royalties from the book *Early Detection and Early Intervention in Developmental Motor Disorders: from Neuroscience to Participation*, published by Mac Keith Press, London; payment or honoraria for lectures; and support for travel to attend international meetings. All the other authors declare no competing interests.

**Bolajoko O Olusanya, Adrian C Davis, Mijna Hadders-Algra, Scott M Wright*

bolajoko.olusanya@uclmail.net

Centre for Healthy Start Initiative, Lagos 101223, Nigeria (BOO); Department of Population Health Science, London School of Economics, London, UK (ACD); Vision and Eye Research Institute, School of Medicine Anglia Ruskin University, Cambridge, UK (ACD); University of Groningen, University Medical Center Groningen, Department of Pediatrics, Division of Developmental Neurology and University of Groningen, Faculty of Theology and Religious Studies, Groningen, Netherlands (MH-A); Division of General Internal Medicine, Department of Medicine, School of Medicine, Johns Hopkins Medicine, Baltimore, MD, USA (SMW)

- 1 Olusanya BO, Boo NY, Nair MKC, et al. Accelerating progress on early childhood development for children under 5 years with disabilities by 2030. *Lancet Glob Health* 2022; **10**: e438–44.
- 2 UNICEF. Seen, counted, included: using data to shed light on the well-being of children with disabilities. 2021. <https://data.unicef.org/resources/children-with-disabilities-report-2021/> (accessed Nov 10, 2022).
- 3 Olusanya BO, Kancherla V, Shaheen A, Ogbo FA, Davis AC. Global and regional prevalence of disabilities among children and adolescents: analysis of findings from global health databases. *Front. Public Health*. **10**: 977453.
- 4 Magnusson D, Sweeney F, Landry M. Provision of rehabilitation services for children with disabilities living in low- and middle-income countries: a scoping review. *Disabil Rehabil* 2019; **41**: 861–68.
- 5 Olusanya BO, Boo NY, de Camargo OK, Hadders-Algra M, Wertlieb D, Davis AC. Child health, inclusive education and development. *Bull World Health Organ* 2022; **100**: 459–61.
- 6 Institute for Health Metrics and Evaluation. Financing global health visualization. 2021. <http://vizhub.healthdata.org/figh/> (accessed Nov 10, 2022).
- 7 Cieza A, Kamenov K, Sanchez MG, et al. Burden of disability in children and adolescents must be integrated into the global health agenda. *BMJ* 2021; **372**: n9.
- 8 The World Bank. Childcare Incentive Fund. <https://www.worldbank.org/en/topic/education/brief/childcare-incentive-fund> (accessed Nov 10, 2022).
- 9 UNICEF. School readiness: a conceptual framework. New York, NY: United Nations Children's Fund, 2012.
- 10 Sapiets SJ, Totsika V, Hastings RP. Factors influencing access to early intervention for families of children with developmental disabilities: a narrative review. *J Appl Res Intellect Disabil* 2021; **34**: 695–711.
- 11 Smythe T, Adelson JD, Polack S. Systematic review of interventions for reducing stigma experienced by children with disabilities and their families in low- and middle-income countries: state of the evidence. *Trop Med Int Health* 2020; **25**: 508–24.
- 12 Thornicroft G, Sunkel C, Alikhon Aliev A, et al. The *Lancet* Commission on ending stigma and discrimination in mental health. *Lancet* 2022; **400**: 1438–80.
- 13 Tordrup D, Smith R, Kamenov K, Bertram MY, Green N, Chadha S. Global return on investment and cost-effectiveness of WHO's HEAR interventions for hearing loss: a modelling study. *Lancet Glob Health* 2022; **10**: e52–62.
- 14 Shahat ARS, Greco G. The economic costs of childhood disability: a literature review. *Int J Environ Res Public Health* 2021; **18**: 3531.
- 15 Banks LM, Polack S. The economic costs of exclusion and gains of inclusion of people with disabilities: evidence from low and middle income countries. 2014. https://www.cbm.org/fileadmin/user_upload/Publications/Costs-of-Exclusion-and-Gains-of-Inclusion-Report.pdf (accessed Nov 10, 2022).