Collectively, through the articles in this supplement, the authors identify numerous challenges to advancing the science of childhood screening but also note exciting opportunities for future research, including potential advances in terminology, theoretical frameworks, and methodologic approaches. The authors’ contributions are grounded in the existing standards of pediatric research but enhanced through contributions from developmental science, econometrics, data science, and public health scholarship. Implementation of the novel and rigorous approaches described herein (and the development of new, yet unimagined innovations) may hold promise for identifying approaches to surveillance and screening that will be the most effective in improving children’s health, development, and well-being. The term “screening” is used differently across various disciplines and research contexts. In the context of prevention, screening is defined as the systematic testing of asymptomatic individuals to identify people at an increased risk (or early stage) of a disease or condition, with the aim of providing information and other services that reduce the risk of complications. Although this definition is relatively straightforward, quantifying the benefits of such screening in clinical practice can be a challenge because of factors such as lack of consistent follow-up diagnostic evaluation and treatment, even in clinical trials. One proposition to address this issue is to expand the definition of screening from mere receipt of a screening test to the entire process from invitation to screening among asymptomatic people through completion of the treatment of the people identified with the condition. Accessible follow-up thus becomes an important issue to consider if the benefits of screening, diagnosis, and treatment are to be quantified at a population level.

“Surveillance” is another term that has different connotations in different contexts. In the public health context, surveillance is generally not considered to be an intervention but, rather, an ongoing system for collecting, analyzing, and using information to assess disease risk and prevalence in a population. Within the United States, surveillance testing at the population level conducted to monitor for disease prevalence may stimulate prevention and control efforts but may not explicitly be linked to follow-up and treatment. In pediatric clinical settings, however, the term “surveillance” has also been used to...
refer to active monitoring at the individual level, which can trigger referrals or interventions. Innovative methods for assessing the impact of surveillance and screening, in all their various forms, are needed to understand and improve their contributions to improved child health outcomes.

Gardner et al illustrate one such approach for assessing the impact of screening using an empirical demonstration of a simulation model. They outline the qualitative dimensions of the screening event and subsequent treatment process that must hold for interventions to be effective: acceptability, accessibility, and fidelity. Their goal is to show impact at the level of population health. Their results reveal that even the best of screening interventions are vulnerable to failures at each step in the health care delivery system. They call for greater integration both within sectors of health care and between health care providers and community contexts, such as schools. Schools and primary care providers complement each other; the former setting is accessible to children whose parents lack health insurance or other ways to pay for medical visits, whereas the latter setting can reach teenagers who have reduced engagement with or have dropped out of formal education.

Building on a tradition of bringing Bronfenbrenner’s ecological systems theory into health research, Graif et al outline a 5-stage socio-ecological model for pediatric screening. Their 5 stages situate individual children within broadening contexts from the microsystems of families, social networks, and health care providers to the macrosystems of populations and public policy. They highlight the selection that can occur within each of these levels, often placing children on pathways to different outcomes. These selective forces, often shaped by experiences of socioeconomic disadvantage, can lead to disparities in access to the preventive care visits in which important screenings occur. These selective forces can also directly influence health outcomes, regardless of whether certain screenings are delivered. Another consequence of selection is that the same behavior may be labeled as within the normative range for members of one group and pathologized for members of another. Taken together, these factors may lead to either overestimates or underestimates of the impact of screening on health outcomes of interest. Better strategies for understanding and accounting for these socio-ecological forces are needed to improve the implementation of childhood screening and translate screening to improved health outcomes.

In her article, Wallis describes how these disparities play out in the specific example of autism screening. Diagnostic algorithms for autism screening were developed largely on the basis of evidence generated in homogenous samples of children. As a result, they may imperfectly predict the risk for children from underrepresented groups. The potential impact of this lack of representation in foundational studies is magnified among low-income children who experience discontinuities in primary care. In these cases, electronic health record evidence of screening and the management of screening results may be missing, whether because of a lack of preventive care, the dispersion of data among multiple providers, or the use of low-resourced providers who are unable to maintain electronic health records. Thus, even when children from underrepresented groups are included in studies, they may contribute proportionally less data, particularly data that can link the delivery of screening to subsequent health outcomes.

One solution noted by Wallis is to consider additional systems with regular contact with children. Her proposal to draw on child care settings dovetails with the socio-ecological model. Teachers and other early education professionals have daily interactions with young children and their families; they also regularly observe young children’s interpersonal skills with other children and caregivers. This microsystem is, therefore, not only a context through which to understand children’s behavioral health but also a source of data that can contribute to the evidence base for a variety of screening recommendations.

In their article, Graif et al also call for community partnerships and data exchanges. Wu et al expand on this idea of “Moving Beyond the Clinic to the Community.” They call for a mesosystems approach of linking across the contexts of families, schools, neighborhoods, and parents’ workplaces through a combination of individual surveys, dyadic data, and administrative records. Community- and systems-level longitudinal linkages create the potential for modeling shared risks in different communities on the basis of webs of connectedness. Social media and genomic data present further sources of currently untapped potential. Such blending of multiple data sources can be done to provide a more holistic and meaningful portrait of key health, developmental, and functional outcomes.

Kemper et al focus on another aspect of the ecological model, the passage of time. They acknowledge...
that considering each screening event in isolation, without consideration of the child's history or context, may be easiest to implement in busy clinical settings and analyze for research purposes. However, this oversimplified approach fails to consider critical factors, such as the complexities of human development and relationships between coexisting health conditions, that can influence screening results but also suggest the most appropriate intervention for a particular individual and setting. Innovative approaches to generating and analyzing longitudinal data and for linking such data to key outcomes are needed accelerate the science of clinical preventive service delivery. Their arguments further explicate the calls from Wallis9 and Wu et al10 for creative approaches to identifying data sources and analytic strategies.

Another component of a developmentally informed approach for new models is to consider novel framing of outcomes. Thus, Silverstein et al12 lay out the case for assessing positively framed outcomes, such as subjective well-being, in addition to traditional negatively framed clinical outcomes, such as the absence of a disease or symptoms. This perspective may tap into measures that hold greater meaning for children and families. To children in elementary school, the ability to hear frequencies across the full spectrum of human vocalization is arguably far less important than their ability to hear well enough to make friends and follow instructions in the classroom. For their parents, an outcome of interest might be hearing well enough to make the required academic progress for on-time promotion to the next grade. These school-aged outcomes occur in much closer time proximity to the recommended developmental screenings for infants and toddlers than commonly used adult health measures do and, thus, may have greater utility for revealing the effects of screening-associated interventions in randomized clinical trials. Moving this area forward may require translational collaborations between developmental psychologists conducting basic behavioral science research and applied work by physician-scientists and public health researchers.

Brown and Kemper13 take this call for rethinking outcomes one step further, by challenging the field to consider more broadly who may benefit from screening, rather than simply what is being measured. They question the core screening principle of limiting consideration of benefit to that which is direct to the patient. Benefits may also accrue to other individuals across the ecological contexts in which children are embedded: family members, peers (such as classmates), and future coworkers and neighbors in the broader community. Economist James Heckman14 has written for decades about the measurable benefit to society of early investment in children's cognitive and noncognitive skills. By an extension of this reasoning, screening for lead in one child that results in the prevention of exposure to lead for a sibling yields a benefit to society by preventing decline in the other child's cognitive and noncognitive skills. Connecting screening to a public health framework also incorporates the consideration of justice, whereby it is ethical to screen a child who will not receive a direct benefit if the harms to that child are minimal and benefits to others are maximized.

Finally, Grosse et al15 describe the complexities of conducting cost-effectiveness analyses of screening interventions in childhood that may not reveal impacts for many years. In their work in econometrics, Heckman et al16 also provide examples of methodologic techniques that may be applied to pediatric prevention. For example, they combine bootstrapping and kernel matching imputation to estimate the societal rate-of-return and benefit-to-cost ratios of a preschool intervention program targeted at young children from families experiencing socioeconomic disadvantage. These methods allow projections to populations beyond those directly observed in the data and across decades of life after the intervention occurs.

CONCLUSIONS

Although there are many methodologic challenges to creating the evidence base for pediatric screening, the opportunities cited above are relevant to multiple stakeholders: funders (e.g., National Institutes of Health [NIH]), researchers, practitioners, health care systems, other systems that serve children (such as schools), public health officials, and policymakers. One effort in which these groups could collaborate is data sharing to create larger data systems. The NIH has invested deeply in data sharing policies for funding recipients17 and sharing the agency's own resources18 as well as supporting efforts in the related areas of data science19 and open science.20 The 5-stage socio-ecological model can provide theoretical grounding for the development of such systems through its attention to the multiple contextual dimensions of screening.

In these articles, the authors also highlight the scientific potential in innovative methodologic techniques and study designs. The development and use of subjective well-being measures that are validated for children, adolescents, and families
hold promise for moving the evidence base forward beyond viewing health as merely the absence of disease. Innovative study designs and analytic methods (such as accelerated longitudinal designs, synthetic cohorts, or regression discontinuity designs) can be used to reduce the need to rely on randomized controlled trials that require extended durations of follow-up before results about the long-term impact of certain exposures or interventions are reported. Econometric techniques applied through approaches such as cost-benefit and cost-effectiveness analyses may address issues of the lack of data to study causal inference over long periods of time by incorporating simulation modeling, big data and data analytics, and natural experiments. In addition, simulation models can be used to help policy makers evaluate whether screening should be provided more broadly and in alternative settings when resources for conducting pilot and implementation studies are limited.

A final important use of the sociocultural model is to bring in the social determinants of health, including structural racism. The NIH offers research resources for the inclusion of the social determinants of health, such as data collection protocols. Along with sister agencies in the US Department of Health and Human Services, the NIH is also soliciting input from the scientific community about ameliorating structural racism in the conduct of scientific research. For example, a recent request for information issued by the Agency for Healthcare Research and Quality called for specific attention to the introduction of racial and/or ethnic bias by the clinical algorithms that shape medical decision-making and the provision of care. This ongoing scientific enterprise dedicated to the generation, improvement, and implementation of the evidence base and resulting recommendations holds considerable potential for generating knowledge that can lead to better health outcomes during childhood and across the life course.

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ABBREVIATION

NIH: National Institutes of Health

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**Challenges and Opportunities to Bolster the Effectiveness of Childhood Screening**

Rosalind B. King, Elizabeth Neilson and Tracy M. King

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