Is a Positive Developmental-Behavioral Screening Score Sufficient to Justify Referral? A Review of Evidence and Theory

R. Christopher Sheldrick, PhD; Daryl Garfinkel, BA

From Developmental-Behavioral Pediatrics, Floating Hospital for Children, Tufts Medical Center, Boston, Mass

The authors have no conflicts of interest to disclose.

Address correspondence to R. Christopher Sheldrick, PhD, 800 Washington St, Box 854, Boston, MA 02111 (e-mail: rsheldrick@tuftsmedicalcenter.org).

Received for publication October 11, 2016; accepted January 26, 2017.

ABSTRACT

In their recommendations on screening for autism and developmental disabilities, the American Academy of Pediatrics recommends referral subsequent to a positive screening result. In this article, we argue that positive screening results are not always sufficient to justify a referral. We show that although positive predictive values are often low, they actually overstate the probability of having a disorder for many children who screen positive. Moreover, recommended screening thresholds are seldom set to ensure that the benefits of referral will equal or exceed the costs and risk of harm, which is a necessary condition for an optimal threshold in decision analysis. Drawing on recent recommendations for the Institute of Medicine/National Academy of Medicine, we discuss the implications of this argument for pediatric policy, education, and practice. In particular, we recommend that screening policies be revised to ensure that the costs and benefits of actions recommended in the event of a positive screen are appropriate to the screening threshold. We recommend greater focus on clinical decision-making in the education of physicians, including shared decision-making with patients and their families. Finally, we recommend broadening the scope of screening research to encompass not only the accuracy of specific screening instruments, but also their ability to improve decision-making in the context of systems of care.

KEYWORDS: autistic disorder; decision-making; developmental disabilities; mass screening; sensitivity and specificity

ACADEMIC PEDIATRICS 2017;17:464–470

WHAT’S NEW

We argue that professional guidelines that recommend referral subsequent to positive results on developmental-behavioral screening instruments should be revised because: 1) positive predictive value often overstates the probability of disorder; and 2) positive scores do not ensure that the benefits of referral outweigh the risks.

The Council on Children with Disabilities and the Bright Futures Steering Committee of the American Academy of Pediatrics (AAP) recommends that pediatricians conduct developmental screening at well-child visits and address positive screening results by making referrals for “developmental and medical evaluations and early developmental intervention/early childhood services.” Many other groups, including the Centers for Disease Control and Prevention, base their recommendations for managing positive screening results on the AAP’s policy statement. In this article, we focus on evidence and theory supporting this recommendation.

On the surface, a recommendation to refer subsequent to a positive result on a developmental-behavioral screening instrument reflects sound judgment. A range of evidence suggests that developmental-behavioral disabilities (a term we use broadly to include developmental delays, autism, internalizing disorders, and externalizing disorders) are underidentified in pediatrics, with approximately a third of children with disabilities being identified in primary care settings. In response, professional guidelines typically recommend use of screening instruments that detect at least 70% of children with developmental-behavioral disabilities (ie, sensitivity = 70%). If these assumptions are true and if all pediatricians began to refer all children who scored positive on valid screening instruments, then detection of developmental-behavioral disabilities at a sufficiently young age could be expected to increase dramatically (ie, from approximately a third of children with developmental-behavioral disabilities to at least 70%).

However, published evidence clearly shows that pediatricians typically do not refer all children who screen positive. A recent analysis of a systematic review identified 8 published studies on the implementation of developmental-behavioral screening programs that reported rates of referral subsequent to positive screens. Among these
studies, referral rates ranged from 10% to 86%. Whereas results such as these are widely interpreted as failures to implement evidence-based screening protocols with fidelity, we suggest an alternative explanation on the basis of at least 2 limitations in the logic underlying many screening recommendations. First, pediatricians make clinical decisions about patients as individuals, one at a time—they do not make decisions for groups of patients. We therefore begin by showing how reliance on group-level statistics like positive predictive value (PPV) overstates the value of recommended screening thresholds (also known as “cut scores”) for making decisions about individual patients. Second, we argue that recommended screening thresholds typically fail to account for the expected costs and benefits of available referral options. Although pediatricians are clearly aware of the benefits of identifying and referring children with developmental-behavioral disabilities so that they can receive treatment, they are also acutely aware of the costs and potential for harm, such as those from false positive results. It is therefore reasonable to hypothesize that pediatricians often do not refer all children who score positive on screening instruments because they perceive the expected costs and harms resulting from referrals to outweigh the benefits, especially when the probability of disability is low. In short, it is reasonable that some children with positive screening results might not be referred. We present each of these arguments in greater detail and conclude by discussing the implications of our argument for pediatric research, education, and policy—including our suggestion that standard recommendations be revised.

**OVERSTATED VALUES**

Group-level statistics like PPV overstate the value of recommended screening thresholds. Screening instruments are often evaluated according to the proportion of children with disabilities who score positive (ie, sensitivity), and the proportion of children without disabilities who score negative (ie, specificity). Many guidelines—including the AAP’s—recommend minimum values of 70% for sensitivity as well as specificity. Statistics like sensitivity and specificity are extremely useful for addressing problems in public health because they set expectations for how a screening instrument will perform when used to identify children with disabilities over an entire population. They are also often used to determine screening thresholds (ie, using the Youden index) to identify the threshold with the greatest combined value of sensitivity and specificity. However, their clinical utility is more limited.

When seeing patients, clinicians must interpret and act on screening results. To inform decisions about referrals for further evaluation or treatment, screening instruments would ideally tell pediatricians the chance that a given child truly has a developmental-behavioral disability. A statistic known as PPV is helpful in this regard. PPV is the probability of having a diagnosis with a positive score. Concretely, it is measured by analyzing all children who score positive and by determining the proportion of children who have a diagnosis. Because it is essentially an indicator of prevalence among the subgroup of children who score positive, PPV is highly sensitive to prevalence overall. If clinicians were to refer all children who scored positive, PPV would reflect how often their referral decisions would be correct using diagnoses as a criterion.

However, as a group-level statistic, PPV suffers from a significant limitation with respect to clinical decision-making: not every child who scores positive on a screening

![Figure 1. Visualization of sensitivity, positive predictive value (PPV), and threshold probability. (A) Normal distributions of screening scores. (B) Sensitivity and PPV: calculated from all true positive (TP), false positive (FP), or false negative (FN) results. (C) Threshold probability: calculated on the basis of frequency (f) of children from healthy and affected populations who score at the threshold.](image-url)
دریافت فوری متن کامل مقاله

امکان دانلود نسخه تمام متن مقالات انگلیسی
امکان دانلود نسخه ترجمه شده مقالات
پذیرش سفارش ترجمه تخصصی
امکان جستجو در آرشیو جامعی از صدها موضوع و هزاران مقاله
امکان دانلود رایگان ۲ صفحه اول هر مقاله
امکان پرداخت اینترنتی با کلیه کارت های عضو شتاب
دانلود فوری مقاله پس از پرداخت آنلاین
پشتیبانی کامل خرید با بهره مندی از سیستم هوشمند رهگیری سفارشات