A Statewide Tiered System for Screening and Diagnosis of Autism Spectrum Disorder

Rebecca McNally Keehn, PhD, HSPP,[1] Mary Ciccarelli, MD,[1] Dorota Szczepaniak, MD, [1] Angela Tomlin, PhD, [1] Thomas Lock, MD[1] & Nancy Swigonski, MD, MPH, MBA [1,2]

Affiliations: [1]Department of Pediatrics, Indiana University School of Medicine, Indianapolis, IN [2]Fairbanks School of Public Health, Indiana University, Indianapolis, IN

Address correspondence to: Rebecca McNally Keehn, Department of Pediatrics, Indiana University School of Medicine, 1002 Wishard Blvd. Indianapolis, IN 46202 Phone: (317) 274-2121 Fax: (317) 948-0126 Email: mcnallyr@iu.edu

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Abbreviations: Ages and Stages Questionnaire (ASQ-3); Autism Diagnostic Interview-Revised (ADI-R); Autism Diagnostic Observation Schedule-Second Edition (ADOS-2); autism spectrum disorder (ASD); Early Autism Evaluation (EAE); Global Developmental Delay (GDD); Modified Checklist for Autism in Toddlers, Revised with Follow-Up (MCHAT-R/F); primary care provider (PCP); Screening Tool for Autism in Toddlers and Young Children (STAT)

Table of Contents Summary: Through development of a tiered system of developmental screening and evaluation, over 2000 young children were evaluated for ASD in the primary care setting.

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Contributors' Statement Page

Drs. Swigonski, Ciccarelli, and Lock conceptualized and designed the study, contributed to designing the data collection instruments, data collection, analysis, and interpretation, and reviewed and revised the manuscript for important intellectual content.

Dr. McNally Keehn contributed to designing the data collection instruments, led the data analysis and interpretation efforts, drafted the initial manuscript, and reviewed and revised the manuscript.

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Abstract

While autism spectrum disorder (ASD) can be reliably detected in the second year of life, the average age of diagnosis is 4 to 5 years. Limitations in access to timely ASD diagnostic evaluations delay enrollment in interventions known to improve developmental outcome. As such, developing and testing streamlined methods for early ASD diagnosis is a public health and research priority. This report describes the Early Autism Evaluation (EAE) Hub system, a statewide initiative for ASD screening and diagnosis in the primary care setting. Development of the EAE Hub system involved geographically targeted provision of developmental screening technical assistance to primary care, community outreach, and training primary care clinicians in ASD evaluation. EAE Hubs implemented a standard clinical pathway for evaluation of children, ages 18-48 months, at risk for ASD. From 2012-2018, 2076 children were evaluated (mean age: 30 months; median evaluation wait time: 62 days) and 33% of children received a diagnosis of ASD. Our findings suggest that developing a tiered system of developmental screening and early ASD evaluation is feasible in a geographic region facing healthcare access problems. Through targeted delivery of education, outreach, and intensive practice-based training, large numbers of young children at risk for ASD can be identified, referred, and evaluated in the local primary care setting. The EAE Hub model has potential for dissemination to other states facing similar neurodevelopmental health care system burdens. Implementation lessons learned and key system successes, challenges, and future directions are reviewed.

Autism spectrum disorder (ASD) is a complex neurodevelopmental disability characterized by impairments in social communication and the presence of restricted and repetitive behaviors affecting 1 in 54 children[1] with lifetime costs exceeding $2.4 million.[2] Measurable behavioral symptoms emerge in the first year of life[3-5] and the diagnostic phenotype becomes largely stable around 14 months.[6] Yet, nationally the average age of ASD diagnosis is 4 to 5 years,[7,8] with children from lower income, minority, and rural backgrounds lagging further behind.[9-11] A shortage of expert evaluators, time-intensive evaluations, reimbursement constraints, and provider hesitancy[12] contribute to delays in referral and long evaluation wait times. The significant delay between the emergence of ASD symptoms and diagnosis means that young children are missing opportunities for intervention at the time of optimal neuroplasticity.[13] Accordingly, developing and testing streamlined methods for early ASD diagnosis is a public health and research priority.[7,14]

One important but recently debated[15] method for early ASD detection is universal screening at 18- and 24-months of age. While the American Academy of Pediatrics recommends both universal developmental[16] and ASD[17] screening, the US Preventative Task Force found insufficient supportive evidence.[18] Despite varied results regarding the accuracy of ASD screening,[19-21] evidence indicates that the mean time to diagnosis is significantly shorter for those who do screen positive for ASD,[19] highlighting the importance of maintaining this standard until more reliable measures are developed.

A second strategy for decreasing the age of ASD diagnosis is to improve access to diagnostic evaluations. The field has seen an emergence of promising research on training primary care providers,[22-25] embedding behavioral health providers in primary care,[26,27] and using telemedicine-based diagnostic procedures.[28,29] Many studies employ an evaluation model

where diagnosis is based on developmental history as well as administration of the Screening Tool for Autism in Toddlers and Young Children (STAT),[30] a Level 2 ASD screening measure. Further research is needed to determine the feasibility of scaling this approach to larger systems.

To address the significant neurodevelopmental needs of young children across the state of Indiana, we developed an innovative tiered system of developmental screening and diagnostic evaluation. Our goal was to improve access to early ASD evaluation in children's local communities and support enrollment into evidence-based interventions. This report describes the development and scale-up of the statewide Early Autism Evaluation (EAE) Hub system, as well as outcomes regarding six years of system implementation and sustainability. Lessons learned and key system successes, challenges, and future directions are offered for other regions that may wish to adopt and expand the EAE Hub model.

SETTING

At the time of initiation of the EAE Hub system, Indiana lagged behind the national average in number of children receiving standard developmental screening, had a higher number of children at-risk for developmental, behavioral, or social delays,[31] and had many counties designated as Medically Underserved Areas[32] (see eTable 1). Reliable state-level data on the average age of ASD diagnosis in Indiana does not exist. However, an internal needs assessment indicated that ASD and developmental delay were the two most prevalent diagnoses served in the neurodevelopmental outpatient clinics of the state's largest pediatric hospital, and that most diagnoses were made after children entered the public school system. Furthermore, this assessment revealed that, similar to nationally reported wait times of 6-12 months,[33,34] Indiana children were waiting an average of 9-12 months from referral to evaluation.

EAE HUB SYSTEM MODEL

The guiding framework of the EAE Hub system is composed of three tiers of service: 1) Children receive standard developmental surveillance and screening *and* ASD screening at primary care well-visits; 2) Children, ages 18-48 months, identified as at-risk for ASD are referred to a local EAE Hub for ASD evaluation and counseling on next step recommendations; and 3) Children with complex or equivocal symptom presentation are referred for comprehensive ASD evaluation at a specialty diagnostic center. A framework of quality improvement, coordination of care, community engagement, and planned co-management with the referring primary care provider (PCP) overlays the system. The primary EAE Hub team included an Executive Director (i.e., academic pediatrician), project coordinator, and practice liaisons. Notably, the team included two parents of children with neurodevelopmental disabilities (including ASD), promoting the importance of family-professional-community partnership in this effort. The development and scale-up of the EAE Hub system was funded by a combination of federal and state grants, philanthropy, and individual contracts with EAE Hub institutions (see eMethods for further information).

Developmental Screening Technical Assistance and Outreach

Developmental screening technical assistance to pediatric and family medicine primary care practices was sequentially targeted around geographic regions as each EAE Hub was developed. A practice liaison and pediatrician visited practices to provide education on 1) standardized developmental and ASD screening procedures following American Academy of Pediatrics policy,[16,17] 2) training on the Ages and Stages Questionnaire (ASQ-3)[35] and Modified Checklist for Autism in Toddlers, Revised with Follow-Up (MCHAT-R/F),[36] as well as kits at no cost, and 3) referral procedures for the local EAE Hub and community services and resources. Follow-up technical assistance occurred as needed. Geographically-focused outreach to

community organizations including early intervention agencies, school corporations, advocacy groups, and regional representatives of state agencies was conducted to provide education on the EAE Hub system and develop partnerships to support children and families.

EAE Hub System Development

The EAE Hub leadership team received individualized and intensive training from the developers of the Screening Tools and Referral Training-Evaluation and Diagnosis (START-ED) model.[25] The objectives of START-ED are to provide primary care pediatricians with a functional and streamlined framework and assessment tools for the accurate diagnosis of young children with ASD. The training included both didactic education on ASD evaluation as well as certification in administration and scoring of the STAT, selected because of its utility in the assessment of toddlers in the primary care setting. This training prepared the EAE Hub leadership team to adapt the START-ED model for the development of the EAE Hub training curriculum and clinical pathway.

The first EAE Hub site was piloted at an academic health center-affiliated pediatric primary care clinic, allowing for refinement of the model and training curricula. Additional EAE Hub sites were selected based upon a two-step process including: 1) an analysis of population distribution to target geographic regions, and 2) selection of pediatric primary care practices in targeted regions with known pediatric champions who were actively engaged in early childhood initiatives. Given the general assumption that pediatricians have more formal expertise and experience in atypical child behavior and development, other types of primary care practices (e.g., family medicine) were not recruited as EAE Hub sites.

The goal was for each EAE Hub to be a clinically and administratively self-sustaining site within the system. EAE Hub sites ranged from large health systems to private pediatric

practices, with commitment from their governing leadership to providing this service in their communities. Individual EAE Hubs negotiated evaluation capacity, payment and revenue, office space and support staff needs, and related issues with their home organization. While there was no formal top-down oversight by the EAE Hub leadership team, consultation and ongoing support was provided to sites through individualized technical assistance and a monthly learning collaborative webinar. The collaborative focused on didactic training, case presentations, and practice-based quality improvement discussions. An annual meeting was held to review quality improvement data, share practice updates, assess system needs and goals, and foster relationships to support sustainability.

Each EAE Hub signed a Memorandum of Understanding to document formal collaboration and agreement to 1) develop a clinical team, including a pediatrician or nurse practitioner (NP) and nurse or medical assistant, ideally with the inclusion of an administrative leader and care coordinator to support follow-up care, 2) participate in EAE Hub training, 3) implement the standard EAE Hub clinical pathway (see Table 1), 4) collect quality indicator data, and 5) participate in the monthly learning collaborative and annual meeting.

EAE Hub Training Curriculum Each EAE Hub, including clinicians and staff, participated in an on-site multi-day intensive training on ASD evaluation. Training was provided by academic faculty in general pediatrics, developmental pediatrics, child psychology/psychiatry, and quality improvement science. The didactic curriculum included education on developmental screening, structured developmental history and interviewing techniques including assessment of DSM-5 ASD symptoms, medical and psychological differential diagnosis and common comorbid concerns, communication skills for delivery of diagnosis, and current evidence regarding ASD interventions. Training on billing/coding and practice quality improvement was

provided to EAE Hub clinicians and pertinent practice staff. Regional community agencies were invited to share local resources and families of children with neurodevelopmental disabilities joined the training as "faculty for the day" to share their experiences with diagnosis and navigating systems and services. Clinical practicum training included in-vivo practice and supervision on all steps of the clinical pathway for up to six evaluations of children with 1) typical development, 2) confirmed diagnosis of ASD, and 3) referral concern for ASD. Training faculty provided learners with written feedback, including ratings of performance during observed practicum sessions. Measures of trainee satisfaction guided revisions of the curriculum over time.

EAE Hub Clinical Pathway The EAE Hub model was developed following the principles of the START-ED model[25] whereby clinicians are provided with training on a standard clinical evaluation protocol and assessment tools for diagnosis of ASD in toddlers. In contrast with standard comprehensive ASD evaluation (i.e., which often includes labor-, training-, and cost-intensive diagnostic tools such as ADOS-2[37] and ADI-R[38]), the EAE Hub clinical pathway specifies a brief evaluation protocol designed to be completed in a 90-minute primary care office visit. Evaluation components include review of ASQ-3 and MCHAT-R/F, diagnostic interview to solicit DSM-5 ASD symptoms and medical history, physical exam, and administration of the STAT (see Table 1). The STAT, a level 2 screening tool originally developed for use in children ages 24-35 months, has been shown to have good psychometric properties[39] (i.e., sensitivity=1.0; specificity=0.85; positive predictive value=0.86; negative predicative value=0.92). Additional research has shown promising utility for an extended age range of 14-47 months.[26,40,41] At EAE Hub system initiation, an age range of 18-42 months was targeted. However, over time the age range was expanded up to 48 months based upon available

STAT guidelines (e.g., including use of alternative age-based scoring procedures[39-42]) as well as clinician feedback regarding comfort and desire to serve a broader group of children for which the standard clinical pathway was appropriate.

EAE Hub Data Collection EAE Hubs collected and submitted de-identified data for each evaluation via standardized paper-based visit summary sheets or direct entry into an online database. To minimize data collection burden on EAE Hubs and ensure HIPAA compliance, individual demographic information was not collected; see eMethods and eTable 1 for county- and state-level demographic information. Data were stored in a secure database and analysis was completed using IBM SPSS Statistics Version 26.

EAE HUB SYSTEM OUTCOMES: 2012-2018

From 2012-2018, the EAE team provided technical assistance on developmental screening to 193 primary care practices composed of 798 clinicians (i.e., 82% physicians; 17% nurse practitioners) and their staff. Outreach efforts also included presentations to 136 community organizations including early intervention agencies (N=31), schools (N=38), autism intervention agencies (N= 7), and local community organizations (N=60). Medical presentations were delivered at 73 events (see Figure 1a). Education and outreach efforts were conducted in 76% of Indiana counties (see Figure 2).

Twelve EAE Hubs were developed in pediatric primary care practices across the state of Indiana (see Figure 2), representing 8 health systems. EAE Hub training was delivered to 90 individuals, including 30 clinicians (i.e., 20 physicians, 10 nurse practitioners) and 53 support staff (i.e., administrators, medical assistants, billing specialists, social workers). Over six years, there was a 92% Hub retention rate; one EAE Hub exited the collaborative due to personnel turnover. Engaging the EAE Hub teams in partnership, intensive training, and monthly

continuing education has supported excellent retention of Hub sites and sustainability of the system.

Over six years, a total 2076 children were evaluated across the EAE Hub system. Of 706 PCPs making referrals, 36% had received developmental screening technical assistance from the EAE Hub team, suggesting that educational efforts spread beyond those who received direct technical assistance. EAE Hub evaluations increased over time as Hubs became established in communities and more sites were added across the system (see Figure 1b). By 2018, EAE Hubs made approximately 72% of expected ASD diagnoses in their respective regions, and 15% of expected ASD diagnoses statewide (see eMethods and eFigure 1).

Thirty-three percent of children evaluated received a diagnosis of ASD (see Table 3). In 8% of cases, evaluation results were equivocal and the EAE Hub clinician was unable to make a definitive determination of ASD diagnostic status. While the EAE Hub model mandated referral to a specialty diagnostic center for equivocal cases, limitations in our capacity for follow-up data collection precluded confirmation that these children received further evaluation, and thus, definitive diagnostic status for this group of children is unknown.

Of all children evaluated (i.e., regardless of ASD status), 72% met diagnostic criteria for global developmental delay (i.e., GDD; defined as delays in $\geq$ 2 developmental domains based on ASQ-3 and/or clinical judgment). Sixteen percent of children met neither criteria for ASD nor GDD; 89% of these children were identified as having one or more developmental, behavioral, or medical concern warranting follow-up or intervention (see eTable 2). Together, these findings suggest that even those not diagnosed with ASD were likely to benefit from evaluation.

Across the EAE Hubs the mean age at evaluation was 30 months (see Table 2), significantly less than the national average of four to five years of age[8] and consistent with

existing reports of community-based diagnostic models.[24-27] Compared to historical 9-12 month wait times estimated across tertiary outpatient clinics, median latency from referral to EAE Hub evaluation (i.e., wait time) was 62 days. This finding of improved access through implementation of ASD evaluation in the primary care setting has been found across several smaller studies.[26-29] Decreasing wait times for evaluation services provided in children's local communities has the important potential of increasing access to early intervention and supportive services.

To determine whether frequency of ASD diagnostic outcome, age at evaluation, and wait time differed across EAE Hub sites a series of exploratory analyses were conducted. For these analyses, 11 of 12 EAE Hubs were included; one EAE Hub developed in 2018 was excluded from analysis given small number of evaluations conducted (n=4). The frequency of ASD diagnosis varied significantly ($p<.001$) across site (see Table 3). There was also a significant difference in age at evaluation ($p<.001$) and wait time ($p<.001$) across site (see Table 2). Wait time at the EAE Hub within IUSM's academic outpatient clinic (i.e., Hub 6) was significantly longer than the average at all other EAE hubs combined ($p<.001$).

Findings regarding differences in number of children diagnosed with ASD, age at evaluation, and wait time across EAE Hub site are notable. Previous research has documented the impact of health care provider knowledge and behavior on referrals for ASD evaluation.[43-47] It is likely that referring PCPs may have variable experience with the heterogeneous ASD phenotype as well as different thresholds of concern that prompt referral, both contributing to differences in age and diagnostic profiles of children evaluated in the EAE Hubs. Additionally, some referring PCPs may use the EAE Hub system more broadly (i.e., for children without clear ASD symptoms but with other developmental/behavioral concerns), thus skewing the number of

children diagnosed with ASD at some sites. Hub-specific factors such as catchment area population size and site capacity are likely to account for variable wait times.

Although the development of the EAE Hub system represents a significant advance in improving access to timely ASD evaluation, such statewide efforts have many challenges and interpretation of our outcomes must be considered in the context of several limitations. First, although our clinical pathway was developed from a well-accepted evaluation protocol[25] and involved intensive practice-based training and performance feedback from experts, there was no independent ASD evaluation from which to evaluate diagnostic accuracy or determine diagnosis for those with equivocal diagnosis. We must also understand how child and family sociocultural factors affect access and outcomes through collection of demographic data. Together, these efforts will be critical in further evaluating the validity of such a statewide system. Further, while more young children are now receiving ASD screening and evaluation in their local communities, we did not have reliable statewide baseline measures from which to evaluate system impact. We also cannot draw conclusions regarding the rate of referral for evaluation for those children who screen positive for ASD, or determine whether earlier diagnosis results in earlier entry into intervention and how this may impact child outcome.

LESSONS LEARNED FROM EAE HUB SYSTEM IMPLEMENTATION

To our knowledge this is the largest published report on the development and implementation of statewide system for early ASD screening and diagnosis to date. We offer lessons learned and key system successes, challenges, and future directions for other regions facing similar neurodevelopmental access issues that may wish to adopt and expand the EAE Hub model.

Develop a Committed Interdisciplinary Planning and Leadership Team

A key ingredient for EAE Hub system success was committed interdisciplinary planning and ongoing leadership. The Department of Pediatrics prioritized this pediatric public health need and dedicated time, resources, and faculty expertise to this effort. An internal needs assessment drove system development planning and an interdisciplinary team of subspecialists, family advocates, and general pediatricians met weekly (at 7AM) to carefully construct and debate a statewide approach to improving access to ASD evaluation. Faculty committed effort above and beyond their existing clinical and academic duties to participate in broad leadership workgroups that determined the scope, process, and funding for system development.

Invest in Local Partnerships at System Start-Up

From the beginning, we aimed to cultivate strong partnerships with EAE Hub clinicians and their organizations, and these relationships have been the foundation of sustainability. Over six years of implementation, one of our most significant insights has been the importance of identifying a pediatric champion (typically an MD in a leadership role) at each Hub site. This champion served to coordinate site-specific EAE Hub services and advocate for the importance of the system at the organization and community level. Further, these champions were invaluable in fueling connections between the EAE Hub, our central leadership team, and local community advocates and organizations. Having relational connections in and across communities is necessary to most effectively support children and their families in accessing needed services.

Maintain System Engagement and Momentum

A crucial ingredient to system engagement and sustainability was fostering collaboration through the learning collaborative. Through these webinars our central leadership team focused on nurturing shared pride and ownership of the system among all EAE Hub teams. Sharing quality improvement data underscored the significant impact that each team and our collective

system made. The webinars also allowed for regular problem solving of issues such as challenging clinical cases, insurance reimbursement, and service navigation. The central leadership team was able to keep a pulse on system quality and management issues that required follow-up.

Develop an Infrastructure for Training and Personnel Management

Providing training in ASD evaluation and ongoing maintenance of skills to a large group of PCPs requires significant investment. Selection of faculty with requisite expertise and funding their time and travel to the EAE Hubs to provide on-site training proved challenging. Over time, EAE Hub clinicians and staff, including those involved in supporting service, billing, and quality improvement efforts, retired from or left their institutions. Turnover in personnel created disruption in system operations and capacity, as well as demands for training new team members. Periodic formal continuing education and re-evaluation of diagnostic accuracy and fidelity to the EAE Hub model is critical for quality assurance. Given time and funding constraints, we were not able to invest in these important efforts from system inception, though we suggest that others who undertake adaptation of this model strive to build and fund this infrastructure from the start.

Develop Processes to Measure Impact

Development of initial system outcome measures was focused around goals of decreasing evaluation wait times and lowering the age of ASD diagnosis. Yet, there are critical downstream impacts that must be measured in order to further understand the significance of this statewide system. For example, understanding whether early diagnosis leads to cascading effects including earlier enrollment in intervention, improved child outcomes, reduced burden on the educational system, and lowered lifetime costs will be critical to further system dissemination and funding.

Collaboration with statewide agencies including Birth to Three programs, public school systems, intervention agencies, and healthcare financing organizations is one method for systematically collecting these types of data.

Support Fiscal Sustainability

There has been inconsistent insurance reimbursement for ASD evaluation services which strains individual EAE Hub organizations. Further, payment for 90-minute EAE Hub evaluations is often lower than what would be provided for a higher number of routine office visits. Clinician productivity and reimbursement requirements vary by organization type, and while primary care clinics set within larger health networks may be able to bear some financial burden in support of addressing a critical pediatric need, this is unlikely to be the case for smaller practices.

An additional hurdle has been navigating significant changes in what insurers deem a valid ASD evaluation. For example, some insurers are now mandating inclusion of specific assessment tools (i.e., ADOS-2) in order to authorize ASD intervention services. Yet, use of these tools requires expert diagnosticians and thus contributes to problems with access. Our central leadership team has worked directly with Medicaid and other insurers to provide education about the EAE Hub system and developed standard documentation regarding the ASD clinical pathway, including evidence for how the model is aligned with recommended standards for evaluation. Standardization of system processes, including adherence to specified formats for evaluation reports and insurance appeals, as well as deepening partnerships with insurers is likely to benefit this continued effort.

Avoid Excess Burden on EAE Hubs

In the current healthcare climate, time and capacity of primary care clinicians and their teams are continuously stretched. For most EAE Hub clinicians, efforts related to system

participation (including service delivery) account for less than 10% of their practice, and as such, they must balance demands from their many competing roles. Participation in this statewide effort without any direct funding (e.g., for additional support staff or indirect costs associated with office space, patient billing, clinician/staff training time, and data collection efforts) creates a significant burden on individual EAE Hub clinicians and their organizations. One potential avenue to reduce burden may be to develop a shared infrastructure of support with a state department of health or similar agency. This partnership could potentially allow for funding to support the work of individual EAE Hubs and build capacity for collection of comprehensive longitudinal outcome data to evaluate system impact. These efforts must be priorities for ensuring sustainability, advancing rigorous system evaluation, and improving pediatric population health.

CONCLUSION

Developing a tiered system of developmental screening and early ASD evaluation is feasible in a geographic region facing significant healthcare access problems. Through targeted delivery of developmental screening technical assistance, community outreach, medical education, and intensive practice-based training, large numbers of young children at risk for ASD can be identified and evaluated in the local primary care setting. Although further rigorous testing of the EAE Hub system is warranted, our findings suggest that this model has potential for further expansion and dissemination to other states facing similar neurodevelopmental health care system burdens. Future directions must include evaluation of diagnostic accuracy of the system, an effort that is in progress, as well as measurement of provider and family satisfaction, child intervention enrollment and outcomes, and cost of implementation.

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Figure 1. a) Developmental screening technical assistance, community outreach, and medical education efforts (2012-2018); b) Early Autism Evaluation Hub completed evaluations (2012-2018); asterisks represent EAE Hubs initiated.

Figure 2. Gray shaded areas depict Indiana counties in which developmental screening technical assistance, community outreach, and/or medical education outreach has occurred; Red stars depict location of EAE Hubs.

Table 1

Components of the Early Autism Evaluation Hub Standard Clinical Pathway: Evaluation procedures and tools implemented during the EAE Hub evaluation

1. Review and/or administration of ASQ-3 and MCHAT-R/F 2. Diagnostic interview, including assessment of DSM-5 ASD criteria and medical history, with caregiver(s) 3. Physical exam 4. Administration of STAT 5. Integration of data including screening measures, developmental history and DSM-5 ASD interview, and STAT results to formulate a clinical diagnosis 6. Diagnostic feedback with caregiver(s), including sharing of clinical recommendations and local resources 7. Dissemination of clinical evaluation report to the PCP, including recommended next steps for care management; further consultative follow-up to the PCP is provided as needed and requested

Note: All evaluation procedures are conducted by the EAE Hub clinician (and team support staff) unless otherwise noted. EAE Hub teams were trained to administer ASQ-3 and MCHAT-R/F as part of the evaluation process; however, if these measures completed within 3 months of the EAE Hub evaluation and provided by the referring PCP, they were not always repeated at the time of evaluation.

Table 2

Age at evaluation and wait time for Early Autism Evaluation Hub evaluations

	Mean	Median	Range	SD
Age (months)				
All EAE Hubs (N = 2059)	30.3	30.0	34	6.7
Hub 1 (n = 428)	29.7	30.0	26	6.8
Hub 2 (n = 84)	28.5	28.0	24	6.9
Hub 3 (n = 183)	30.7	30.0	34	7.1
Hub 4 (n = 102)	30.2	30.0	24	6.8
Hub 5 (n = 151)	30.5	30.0	28	7.1
Hub 6 (n = 464)	30.2	30.0	24	5.4
Hub 7 (n = 389)	32.0	32.0	28	6.6
Hub 8 (n = 150)	29.5	29.0	30	7.2
Hub 9 (n = 22)	30.1	30.5	22	6.5
Hub 10 (n = 31)	27.5	26.0	23	5.6
Hub 11 (n = 55)	28.96	29.0	23	6.3
Wait time (days)				
All EAE Hubs (N = 1674)	76.82	62.0	341	55.9
Hub 1 (n = 403)	65.1	54.0	324	51.4
Hub 2 (n = 13)	30.4	27.0	71	17.4
Hub 3 (n = 94)	37.7	31.5	220	31.0
Hub 4 (n = 43)	61.6	40.0	316	60.5
Hub 5 (n = 142)	62.8	57.0	184	32.1
Hub 6 (n = 410)	122.9	123.5	337	61.6
Hub 7 (n = 366)	72.5	77.0	200	35.9
Hub 8 (n = 143)	42.8	30.0	316	47.0
Hub 9 (n = 16)	91.3	74.0	145	46.5
Hub 10 (n = 29)	48.9	44.0	183	48.9
Hub 11 (n = 15)	32.13	29.0	49	12.7

Note: Total EAE Hub evaluations (2012-2018) = 2076; data reported above reflects some missing values for both age and wait time. It should be noted that wait times were not consistently reported by all EAE Hubs for the duration of the project.

Table 3

Number and percentage of children diagnosed with ASD in the Early Autism Evaluation Hubs

	Number	Percent
ASD Diagnosis		
Total EAE Hub Evaluations (N = 2076)	691	33.3
Hub 1 (n = 429)	128	29.8
Hub 2 (n = 86)	18	20.9
Hub 3 (n = 184)	36	19.6
Hub 4 (n = 85)	39	38.2
Hub 5 (n = 151)	49	32.5
Hub 6 (n = 464)	169	36.4
Hub 7 (n = 389)	147	37.8
Hub 8 (n = 150)	57	38.0
Hub 9 (n = 25)	11	44.0
Hub 10 (n = 31)	8	25.8
Hub 11 (n = 61)	26	42.6

Percent is based on total number of children evaluated in the EAE Hubs from 2012-2018 (N=2076).

Figure 1

Developmental screening, community outreach, and EAE Hub evaluations by year

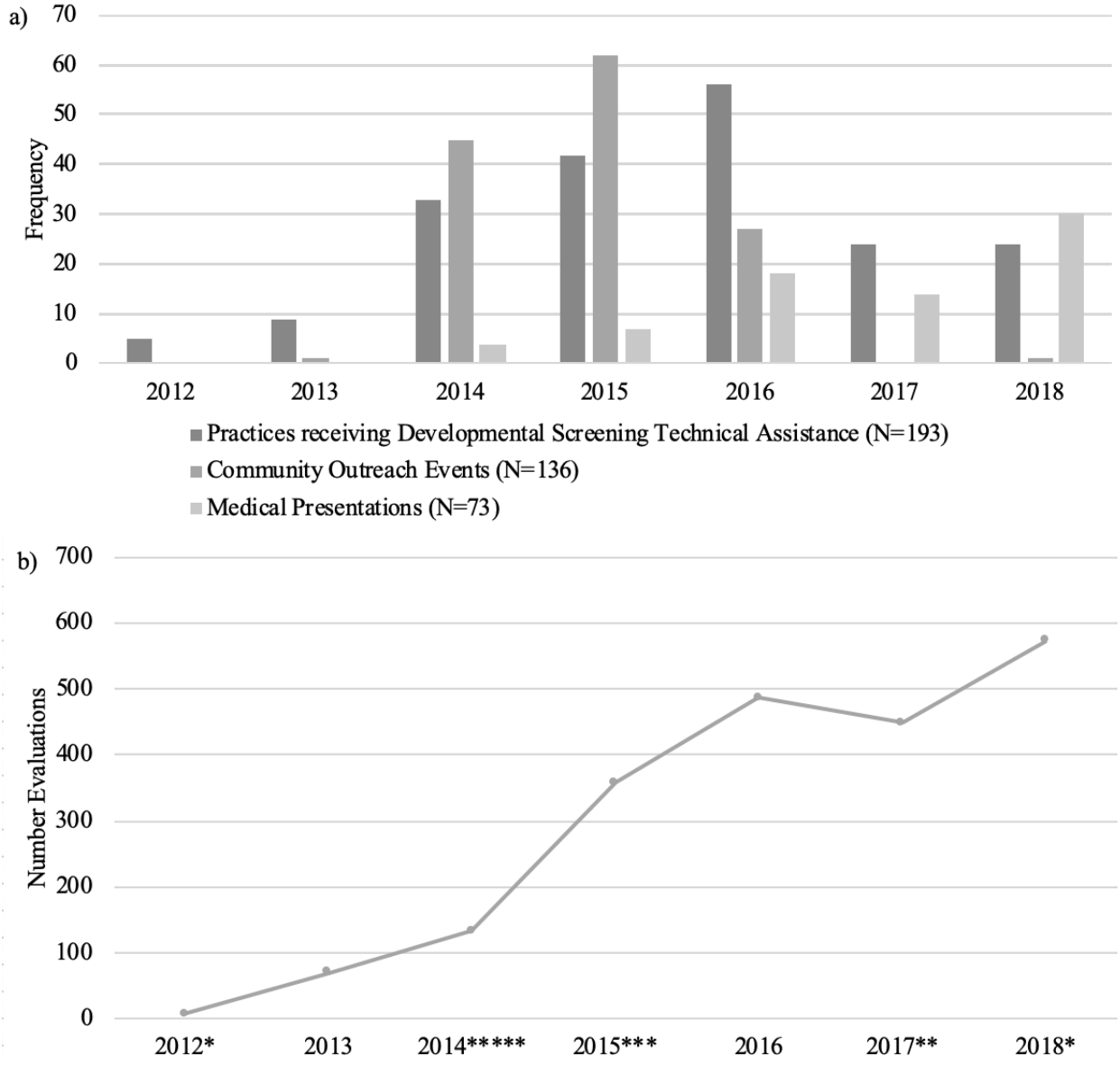
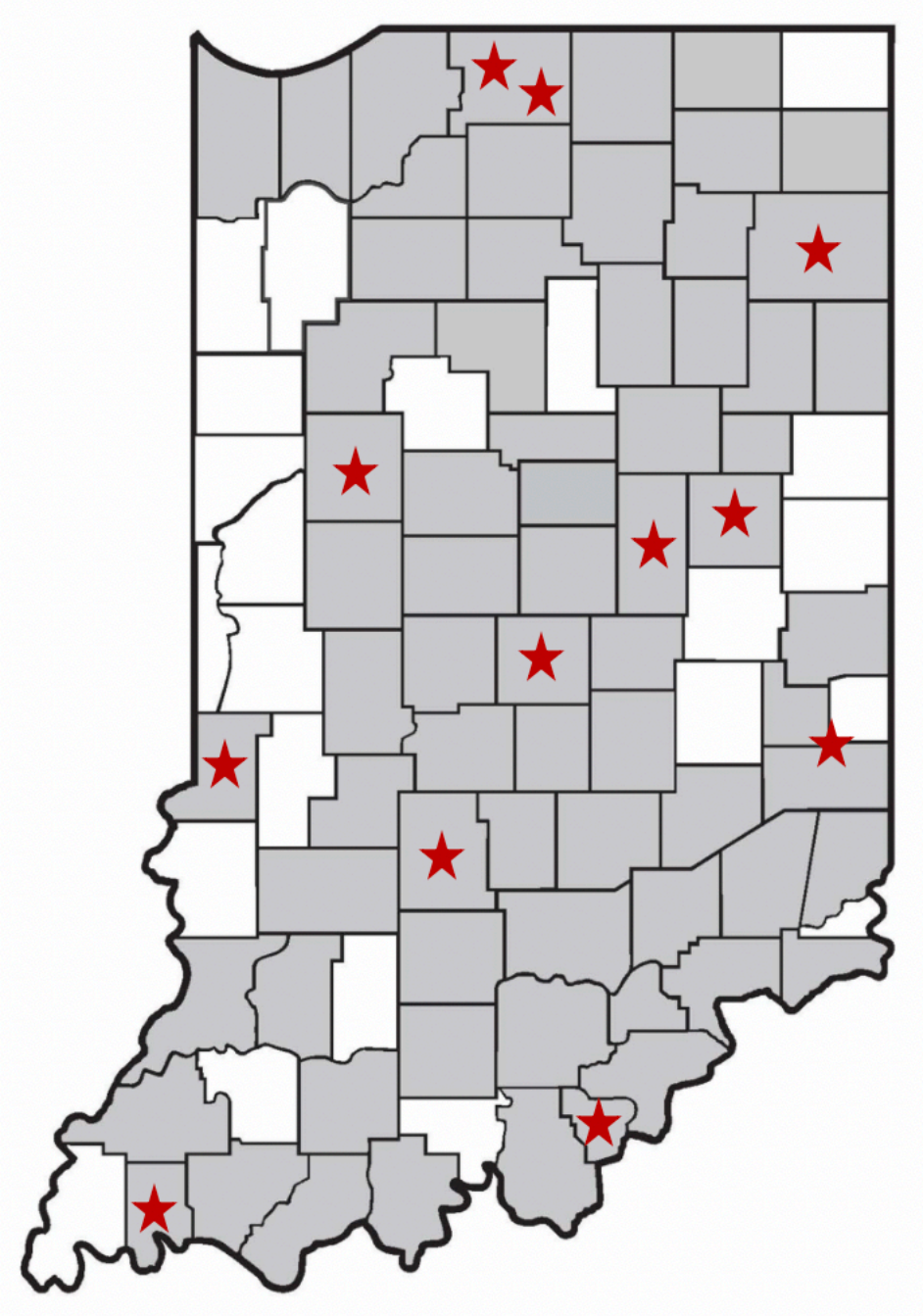


Figure 2

Geographic illustration of community efforts and Early Autism Evaluation Hub installment



Supplementary Materials

eMethods. Supplementary Methods

Indiana statewide and Early Autism Evaluation Hub county demographics and Medically Underserved Area designations

Population-based demographic information for counties in which Early Autism Evaluation (EAE) Hubs are located, as well as for the state of Indiana, are reported in eTable 1. Sex, race, ethnicity, education, and economy and income data was gathered from the U.S. Census Bureau[1] (reporting time period 2014-2018). Additionally, Medically Underserved Area[2] (MUA) designation is reported by EAE Hub county. MUA are federal designations of a geographic area (i.e., county) which meet specific criteria for needing additional primary health care services.

Early Autism Evaluation Hub system cost

The development of the EAE Hub system was funded by a combination of federal and state grants, philanthropy, and individual contracts with EAE Hub institutions. The approximate cost for the academic medical institution to oversee the statewide EAE Hub system was $190,000 per year, which included salary support for the EAE Hub Director (i.e., 0.2 FTE equivalent) and two practice liaisons (i.e., 1.0 FTE each). The practice liaisons were responsible for coordination of all system activities including, but not limited to, statewide developmental screening and educational outreach efforts, EAE hub training, data collection and management, EAE Hub quality improvement initiatives (including Maintenance of Certification activities), and monthly learning collaborative and annual conference. Additionally, regular and ongoing statewide advocacy efforts were required to address problems with billing, reimbursement, and service access that served as barriers to both the EAE Hubs as well as the children and families they served. Each EAE Hub training was estimated to cost approximately $20,000. These funds covered development and individualization of the multi-day training curriculum (including preparation of materials by EAE Hub practice liaisons), scheduling of training faculty, faculty travel to EAE Hubs, scheduling of patients for practicum portion of the training, and coordination of training activities and efforts with the EAE Hub.

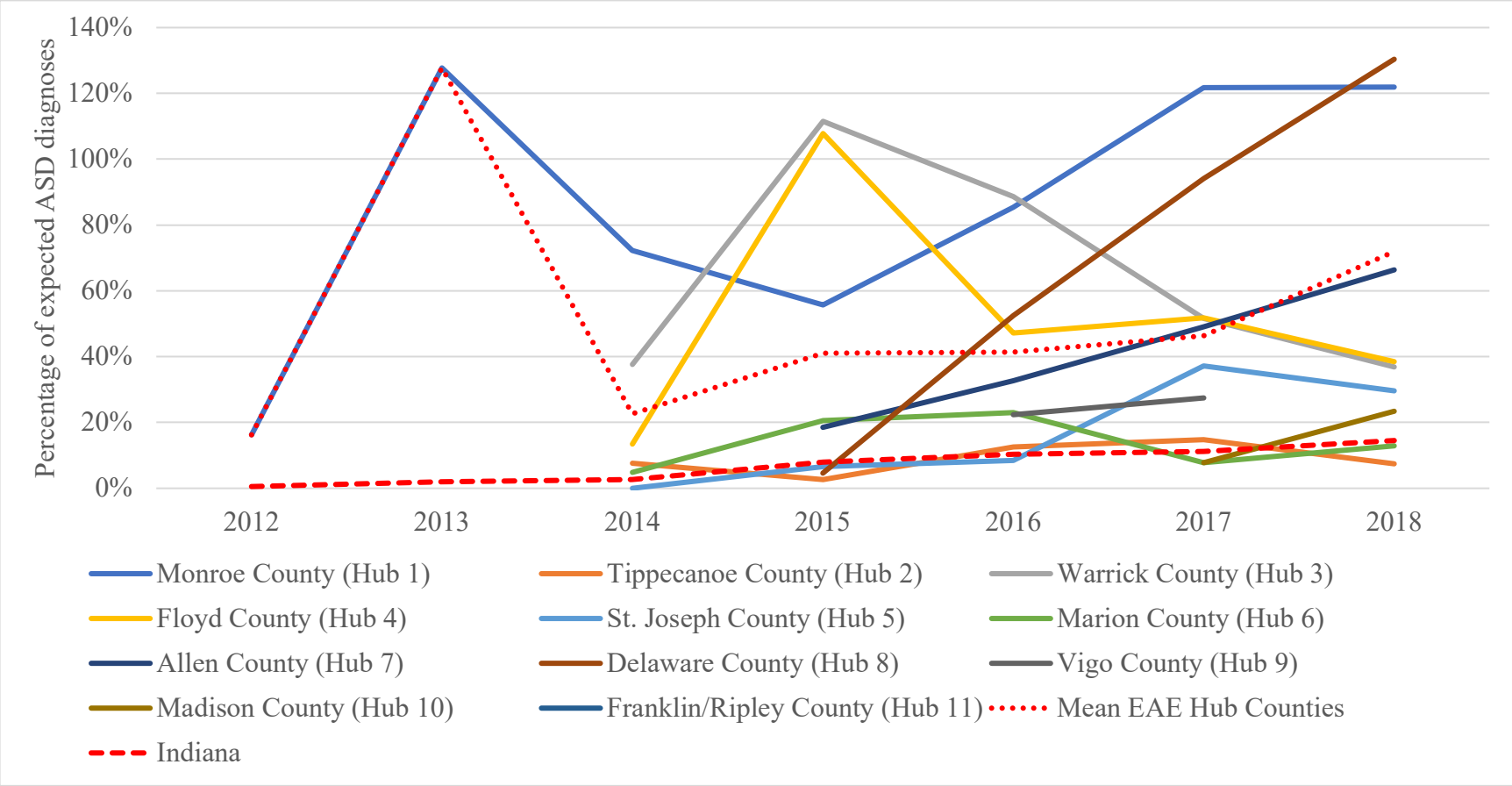
There was no exchange of funds from the leading EAE Hub academic institution to individual EAE Hubs to support system activities. EAE Hubs were fiscally independent from the EAE Hub academic medical institution and were responsible for all costs associated with implementation at their site. EAE Hubs were provided guidance on evaluation billing procedures during initial training and ongoing advocacy from the leadership team regarding billing and reimbursement was conducted. Individual EAE Hub sites subsidized costs of implementation via billing for patient evaluations. Specific data regarding costs of individual implementation of the EAE Hub to the home institutions has not been collected.

Percentage of expected ASD diagnoses made in Early Autism Evaluation Hubs

The number of children diagnosed with ASD in the EAE Hubs (i.e., individually and as a system), as well as across the state of Indiana, was compared with the expected number of ASD diagnoses in that geographic region by year. Indiana birth record data[3] (by county and state) and Center for Disease Control and Prevention (CDC) prevalence rates[4] by year were utilized to calculate estimation of coverage. Specifically, in order to obtain a *general estimate* of the number of expected ASD diagnoses in children ages 18-48 months (mean age at evaluation = 30

months), we utilized birth record data to obtain numbers of children expected to turn two years during the evaluation year (i.e., e.g., 2012 birth rates were used to estimate the number of children turning two in 2014). It is important to note that CDC ASD prevalence data (i.e., 1 in 69 children in 2012; 1 in 59 children in 2014) was estimated based on ASD diagnoses in 8 year old children[5] and that calculated prevalence rates for younger children are lower[6], suggesting that use of the CDC published prevalence rates as a baseline for expected diagnoses in toddlers likely overestimates the expected prevalence for toddlers in our geographic regions.[7] Nevertheless, we provide general estimates regarding the EAE system coverage of expected ASD diagnoses by county and state in eFigure 1.

eFigure 1. Percentage of expected ASD diagnoses made in Early Autism Evaluation Hubs by geographic region (2012 – 2018)



Mean EAE Hub coverage of expected ASD diagnoses in each county increased from 16% in 2012 to 72% in 2018 (see eMethods). Coverage of expected ASD diagnoses varies by EAE Hub, with regional population (i.e., birth rates) and number of EAE Hub providers and site capacity likely influencing these findings. Percentages of expected diagnoses over 100% may be interpreted in context of the following: 1) EAE Hubs receive a portion of referrals from outside their individual county, and 2) in the first one to two years of implementation, individual EAE Hubs have an influx of regional referrals.

eTable 1. Indiana statewide and Early Autism Evaluation Hub county demographics and Medically Underserved Area designations

		Monroe County (Hub 1)	Tippecanoe County (Hub 2)	Warrick County (Hub 3)	Floyd County (Hub 4)	St. Joseph County (Hub 5)	Marion County (Hub 6)	Allen County (Hub 7)	Delaware County (Hub 8)	Vigo County (Hub 9)	Madison County (Hub 10)	Franklin/ Ripley County (Hub 11)	**Indiana**
Sex	Female (%)	50.3	48.9	51	51.4	51.3	51.8	51.1	51.7	49.3	50	50.25	**50.7**
Race	Caucasian (%)	86.4	83.2	93.8	90.5	79.9	64	79.9	88.8	87.9	88.3	97.2	**85.1**
	Black or African American (%)	3.6	5.7	1.7	5.5	13.8	28.9	12	7.2	7.3	8.6	0.4	**9.8**
	American Indian and Alaskan Native (%)	0.3	0.4	0.3	0.4	0.6	0.5	0.5	0.3	0.4	0.4	0.35	**NA**
	Asian (%)	7.0	8.6	2.7	1.2	2.6	3.6	4.4	1.4	2	0.6	0.55	**2.5**
	Native Hawaiian and Other Pacific Islander (%)	0.1	0.1	<0.1	<0.1	0.1	0.1	0.1	0.1	<0.1	0.1	<0.1	**0.1**
	Two or more races (%)	2.6	2.2	1.6	2.4	3.1	2.9	3.2	2.3	2.4	2	1.05	**2.1**
Ethnicity	Hispanic or Latino (%)	3.5	8.5	1.9	3.4	8.9	10.6	7.6	2.6	2.7	4.1	1.55	**7.1**

	Not Hispanic or Latino (%)	83.4	75.4	92.2	87.7	72.2	54.8	73.5	86.7	85.5	84.8	95.9	**78.9**
Education	High school graduate or higher (%)	92.2	91.7	93	90.3	88.4	85.7	89.3	89.3	88.1	88.1	88.8	**88.6**
	Bachelor's degree or higher (%)	45.6	37.7	29.6	29.5	29.2	30.4	28.2	24.3	24.5	18	18.95	**25.9**
Economy/ Income	In civilian labor force (%)	62.1	64.1	66.9	66.1	63.9	67	66.6	58.8	59.8	57.4	64.3	**63.8**
	Median household income ($)	$47,075	$51,844	$70,468	$61,574	$50,938	$46,692	$53,042	$42,705	$43,859	$47,436	$57,338.50	**$54,325**
	Persons in poverty (%)	21.4	18.1	7.1	9.5	13.5	17.2	13.5	22.5	19.2	17	9.85	**13.1**
MUA	X denotes MUA designation	X	X		X	X	X	X	X	X	X	X	

MUA = Medically underserved area (includes geographic area and low-income population designations); NA=data not available

eTable 2. Diagnostic concerns in children without Autism Spectrum Disorder or Global Developmental Delay

Diagnostic concern	N	%
Speech language delay	189	55
Behavior regulation concern	92	27
Sleep problem	37	11
Sensory processing concern	31	9
Other delayed milestones	29	8
Social emotional concern	26	7
Involvement with child protective services/ foster care system	20	6
Motor delay/impairment	22	6
Feeding problem	18	5
Other medical concern requiring specialist follow-up (including lead exposure, genetic concern)	18	5
Concern for seizures	11	3
Toileting problem	7	2
No documented diagnostic concern	**28**	**11**

N=number of children (out of 344) in which a specified (non-ASD/GDD) diagnostic concern was identified in the EAE Hub evaluation; %= percentage of children (out of 344) in which specified diagnostic concern was identified; EAE Hub clinicians were able to report more than one diagnostic concern per child

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