Priorities Established by the Combating Autism Act for Improving ASD Identification: Looking Beyond Ideas and Instruments Towards Implementation

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Background & Objectives

Created in response to growing concerns about ASD's prevalence and impact, the 2006 Combating Autism Act or CAA is the most ambitious ASD national research strategy to date. CAA mandated the InterAgency Autism Coordinating Committee or IACC to set priorities for federal research funding. Between 2008 & 2013, IACC monitored the allocation of $1.8 billion towards these priorities, with more than $1 billion from the US National Institutes of Health or NIH alone (Office of Autism Research Coordination, 2017).

The lack of significant improvement in timely and accurate identification, especially for traditionally underserved groups, has led researchers and advocates to question the heavy emphasis traditionally placed by NIH and other agencies on basic science research. To date, few if any independent researchers have sought to systematically reconsider IACC's priorities and potential impact. Would a greater focus on research that addresses barriers to community-based services, like disparities in access or the challenges of capacity building, be more effective in closing gaps in ASD identification?

Objectives:

- To establish how many NIH-funded projects funded between 2008 and 2013 to address identification of people with ASD sought to directly improve community-based services, close gaps for underserved groups, or build overall system capacity.
- To explore possible reasons for the lack of projects addressing immediate implementation in community settings, beginning with the clinical training and the experience of project leaders with regards to community-based services.

Methods

ARD Database searches

We utilized the Autism Research Database or ARD to identify projects undertaken by NIH and focused on ASD identification. ARD was created and is managed by the Office of Autism Research Coordination or OARC. OARC was created to support the activities of the IACC. ARD is organized around the principle questions identified by the IACC as part of their strategic plan (Office of Autism Research Coordination, 2017). ARD assembles a range of information (project title, principal investigator or PI, abstract, funding agency, funding amount, federal application ID). These data are freely available for download (for 2013 data, click here).

The different sources of funding for ASD projects were assessed in a first phase, through a series of steps that relied initially on broad categories captured in ARD. These are described below, and summarized in Figure 1. In this initial phase, we focused on the funds dispensed. Subsequent stages also include information on the number and types of projects funded, and the characteristics of principal investigators.

1. *Federal*: We identified all projects within ARD that were funded by the federal government, as distinguished from private sources like the Simons Foundation and Autism Speaks.
2. *DHS*: We identified the subset of federal projects and funding initiated by Department of Health and Human Services or DHS. Other federal agencies that initiated a significant number of ASD projects captured in ARD include the Department of Defense, and the Department of Education.
3. *NIH*: We identified the subset of DHS projects and funding captured by ARD and initiated by the NIH, one of the most important divisions within DHS. Other divisions within the DHS that initiated ASD projects captured in the ARD include the Administration for Community Living, the Administration for Children and Families, the Agency for Healthcare Research and Policy, the Centers for Disease Control and Prevention, the Center for Medicare and Medicaid Services, and the Health Services and Resources Administration.
4. *Projects categorized under ASD identification*: We identified ASD projects undertaken by the NIH that were categorized in ARD under Question 1 (Screening and Diagnosis) on the IACC's Strategic Plan.

Defining implementation

Dimensions of implementation. Implementation here is defined as the use of *clinical tools* to target *clinical outcomes* for a *clinical population* in a *community setting*. Each italicized term is described in greater detail below. In general, these definitions draw on a level of detail not always available in the materials published by researchers, and sometimes rely on the judgment and experience of the reviewer. In many cases. these details are intended to exclude initial research studies that might yield findings with implications for assessment or treatment, from studies of tools or techniques could be ready for immediate use by community-based professionals, were training and funding made available.

The use of the term **clinical** throughout does not restrict these definitions to medical settings, methods, or professionals. It is intended to encompass any specialized assessment or intervention activities delivered by, or under the supervision of, any specially trained professional, including those in medical, education, or community settings.

A **community setting** is one that is is typically mandated to provide day-to-day assessment or treatment of ASD. This includes schools, outpatient clinics, community-base behavioral health program, and so on. This also includes any training provided to parents, and services provided in the person's home. It does not include specialized, university-based clinics, or more specialized programs not typically available through regional children's children's hospitals. This distinction is intended to exclude a specialized assessment or treatment program only available through a given children's hospital because of the hospital's role in developing a given research protocol. In such cases, the specialized assessment or treatment program is unlikely to be immediately accessible to other hospital or community settings, even with training and funding. One

exception would be routine ASD diagnosis and assessment, using established and widely available tools, which is commonly available at most children's hospitals.

A **clinical tool** is a specific test, drug. intervention method, or program of services that could be used by a professional in a community setting for purposes of assessment or treatment. This tool is a recognized method for which training might be reasonably obtained through initial licensure, workshops, direct consultation, or materials accessible to the community-based clinician or educator. In many cases, information about the validity, reliability, and likely outcomes have already been published. This definition is intended to exclude a preliminary research finding only indicating a possible relationship between some outcome, and some independent variable suggestive of an intervention. This definition also excludes a method under development and that has yet to be validated. In both such cases, the tool is not reasonably likely to be used with a reasonable level of fidelity by a professional in a community setting.

A **clinical population** is a group of individuals who have been diagnosed with a condition that merits treatment by a professional. This definition is intended to exclude research focusing on participants identified with characteristics of ASD or a related condition, but yet to be formally diagnosed. The exception would be for research exploring the use of a tool to screen for or diagnose ASD.

A **clinical outcome** is a skill or behavior that may realistically be the target of a program of assessment or treatment delivered by a professional. This definition is intended to exclude a preliminary research study first exploring a relationship between an intervention and a very specific characteristic that might not be the target of treatment.

Implementation coding

Coding for Levels of Implementation is summarized in Figure 1. The levels of primary interest to the present study are those relevant to immediate implementation in community settings (e.g., Level III). These codes were intended to capture projects focused on delivering services in community settings, closing gaps for underserved populations, and increasing system capacity. The Pre-Implementation level was intended to capture tools or techniques that might eventually be used in community settings, because they piloted clinical tools with clinical populations in more specialized settings. Within some of these codes, we also distinguished between those projects that assessed clinical tools, and those projects that actually tested their delivery. All other projects were coded as contributing to basic science, including those focused on research infrastructure or the training of researchers. The order of codes was intended to capture increasing level of relevance to large-scale implementation. If a project fell clearly within two codes, we assigned the higher code.

Phase 1 Coding of IACC subcategories for community implementation

We first applied supplemental codes to the IACC subcategories adopted by ARD related to Question 1(Screening and Diagnosis). These subcategories were derived from the IACC Strategic Plan, and summarized in the 2012-2013 Portfolio Analyses (see Office of Autism Research Coordination, 2017, pp. 34). Nine specific subcategories were identified by the IACC within Question 1. A tenth subcategory covers projects generally relevant to the broader question of screening and diagnosis, but which could not be assigned to any of the 9 subcategories.

Phase 2 Coding of individual projects for community implementation

In Phase 2, we conducted reviews of the abstracts of individual projects within those subcategories immediately or eventually relevant to implementation (e.g., Levels II and III). At the time of writing, abstracts awee not easily downloaded from ARD, and so we first downloaded relevant files from NIH REPorter (ARD has since expanded the information downloadable in a single file). The NIH REporter tools provide access to files containing more detailed information about NIH grants, including project abstracts and the publications resulting these projects, as well as the unique identifiers needed to link this information to projects captured in ARD.

Coding project abstracts. These individual project reviews focused on any text in the abstract indicating; (a) the primary or secondary hypotheses, aims, or goals of the study, or (b) the long-term implications of relevance to public health. We also coded long-term implications and implications for public health using the same system for coding level of implementation described above.

We also scanned the titles of other projects within the "Other" subcategory to identify projects with the potential to be immediately or eventually relevant to community implementation. If so, the abstracts of these projects were also reviewed in the same manner as described above. The goal here was to identify the subset of projects that were clearly and immediately relevant to implementation to subject to the review outlined above.

Background of PIs. To explore factors that might explain a relative lack of projects focused on community implementation, we began by considering the related clinical training and community experience of PIs of projects aiming to improve implementation. We selected all of the projects with specific aims or presumed long-term relevance for eventual implementation (e.g., Levels II and III) as identified in the previous step, We downloaded the resumes of PIs through Google© searches. For evidence of clinical training, we scanned resumes for information indicating the completion of a clinical degree and/or licensure in a recognized field of medicine, allied health sciences (e.g., psychology, speech-language pathology, occupational therapy, and so on), or education. For evidence of significant experience in community settings, we scanned resumes for information indicating at least 5 years of work after the completion of all requirements for licensure, delivering relevant services in a community setting, as defined earlier. We also considered evidence indicating experience leading community-based programs of services. Resumes that included a clear timeline of education and work history for at least the past 10 years were retained for coding.

Results

Review of sources for funding ASD projects

A review of projects listed in ARD revealed 6916 projects funded for $1,886,048,017 between 2008 and 2013 (see Figure 2).

1. *Federal Sources*: $1,444,950,169, or 77% of all ASD project funding came from federal sources. Private sources accounted for $441,097,848 or 23% of all ASD project funding. Almost all of this private funding came through the Simons Foundation ($305,632,750 or 16% of total funding) and Autism Speaks ($119,218,935 or 6% of total funding).
2. *DHS*: $1,265,842,089, or 88% of all federal funding for ASD projects was provided through DHS. DHS was responsible for 67% of all funding captured in the ARD

database. Other federal agencies providing significant ASD project funding included the Department of Education ($122,062,861 or 6% of total funding), the Department of Defense ($32,088,323 or 2% of total funding), and the National Science Foundation ($22,539,916 or 1% of total funding).

3. *NIH*: $1,067,409,700, or 84% of all DHS funding for ASD projects was provided through NIH. NIH was responsible for 57% of all funding captured in the ARD database. Other DHS entities providing significant ASD project funding included the Centers for Disease Control ($106,587,127 or 6% of total funding), the Health Services and Resources Administration ($84,044,185 or 4% of total funding), and the Agency for Healthcare Research and Quality ($3,763,606 or less than 1% of total funding).
4. *Projects categorized under ASD Identification*: $167,516,997, or 16% of all NIH funding for ASD projects was categorized in the ARD under Question 1. This funding was directed through a total of 429 grants involving 173 distinct projects. The other questions meritting the most significant level of funding from NIH clearly involved Basic Science, and addressed the biology ($349,483,470, or 33% of all NIH funding) and risk factors ($232,811,921, or 22% of all NIH funding) associated with ASD.

Comparing subcategory funding for NIH versus other agencies. We also summarized the relative emphasis placed on different subcategories by the NIH and by other agencies (see Figure 3). The $167 million spent by the NIH was 3 times more than all other public and private agencies combined. For all agencies, the majority of funding was dedicated to Q 1.L.A (Behavioral and biological markers) and Q 1.L.B (Measures of behavioral / biological heterogeneity). The greatest difference between the NIH and other agencies with respect to funding patterns lay in the relatively greater emphasis placed by the NIH on new diagnostic tools for diverse samples (Q 1.S.A).

Phase 1 Coding of IACC subcategories for implementation

Coding of IACC subcategories for implementation is summarized in Figure 4. Most of the subcategories fell clearly into Basic Science. While projects captured under Q1.L.A (Measures of behavioral and/or biological heterogeneity) may eventually help to identify potential markers, the level of risk would still have to be established before a specific tool is developed. Two of the subcategories meritted coding for Pre-Implementation. While the goal of Q1.S.A (New diagnostic tools for diverse samples) potentially helps to close gaps, it still appeared to focus on the development of new tools rather than their deployment in community settings. Only two subcategories appeared to clearly involve implementation in community settings. In both cases, these appeared likely to focus on assessing needs rather than demonstrating strategies for improvement.

We also summarized the level of NIH funding for subcategories of projects by level of implementation (see Figure 5). The vast majority (66%) of NIH funding related to ASD screening and diagnosis appeared to be dedicated to projects focused on questions of basic science. In this first phase of coding, less than 1% of total funding appeared to be devoted to addressing implementation in community setting; in this case, assessing access for traditionally underserved populations.

Phase 2 Coding of individual projects for implementation

Coding of individual project abstracts and the background of Project PIs is summarized in Figure 6.

Coding project abstracts. A total of 78 project abstracts were reviewed - i.e., abstracts from all IACC subcategories from Question 1 except those categorized as Basic Science. A total of 9 projects were identified with project aims involving implementation of community-based services. The cumulative cost of about $10.1 million was substantially higher than suggested by the original coding of subcategories. This represented about 6% of total funding for Question 1, but only about 1% of total NIH funding.

Coding the background of PIs. A Google© search for the resumes of 9 PIs of projects that included specific aims involving identification in community-based settings yielded 6 resumes. Review of these resumes indicted that the majority (4 PIs, or 67%) were clinically trained, almost always as psychologists or physicians. None of these PIs had significant experience delivering services in community-based programs, let alone leading such programs. The psychologists identified appeared to have all moved directly into faculty positions, sometimes after a brief tenure as a psychologist or postdoctoral fellow on a research project. The physicians identified were more likely to have significant clinical experience, although this appeared to only occur within specialized children's hospitals.

Results and Conclusions

These analyses indicate that relatively few of the projects funded by the NIH between 2008 and 2013 seemed likely to test a specific and immediately applicable tool or approach to improving ASD identification in the community. A systematic review of individual project abstracts originally categorized by ARD as addressing ASD identification revealed that only 6% of the funds ostensibly dedicated to improving screening and diagnosis actually addressed ASD identification outside of specialized university and hospital settings. This represented a negligible proportion of overall NIH funding.

This gap is striking given that the benefits of early identification, combined with rising prevalence estimates, are so often cited as a driving force for ASD research, and were central to the argument for the increased funding provided by CAA. The paucity of research addressing service gaps and barriers severely limits the impact of all other research on ASD identification; even the most powerful tools for screening and diagnosis will have little impact if effective community-based services cannot be developed, gaps in reaching underserved groups cannot be closed, and barriers to building capacity cannot be overcome. In this context, the fact that the average age of diagnosis remained unchanged in the most recent prevalence estimates from the Centers for Disease control is not surprising.

These analyses also reveal that much more funding was spent by NIH on basic research, and somewhat less on projects that piloted tools in specialized settings. Indeed, these findings shed new light on NIH's heavy emphasis on basic science; during this period, it dedicated 55% of funds towards the biology (Question 2) and causes (Question 3) of ASD, and up to an additional 24% towards research training and infrastructure (Question 7). In other words. almost 4/5s of all funding appears to have been dedicated to projects addressing questions of basic science, or supporting the infrastructure needed to ask such questions. Additional reviews of a sample of

individual project titles and abstracts, as conducted here on projects in the Other subcategory, will be helpful in confirming these trends.

Further research will be needed to elucidate possible reasons for the scarcity of projects addressing community implementation of ASD screening and diagnosis. The present analysis suggests that this disinterest may stem from the absence of significant community experience among the PIs reviewed. Such experience can sensitize clinical researchers to the urgency of the need, as well as to specific strategies to close these gaps. PIs who are not clinically trained will always struggle, however, to understand the complexities of identifying ASD quickly and effectively, let alone closing the gap in community implementation. Additional research characterizing the background of all members of the research team may reveal other contributors who can draw on significant community experience.

Additional research may confirm and extend these findings in other important ways. The review of individual project abstracts revealed that the reliance on broad subcategories is inadequate to capture details about the specific aims and presumed relevance of the project. To confirm these trends, abstracts from other subcategories may need to be sampled to evaluate if other projects addressing implementation in community settings are being missed. It is also important to recognize that important project details can never be gleaned from these abstracts alone. By reviewing the resulting publications, for example, we can verify whether the resulting study addressed community implementation. This might allow us to explore whether the findings themselves are likely to impact actual community practice, based on a review of the number, type, and quality of recommendations.

References

Office of Autism Research Coordination. (2017). 2013 he review of individual project abstracts IACC Autism Spectrum Disorder Research Portfolio Analysis Report. Retrieved from https://iacc.hhs.gov/portfolio-analysis/

For further information about this project, please contact
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Figure 1: Coding for levels of Implementation

Terms that are underlined are the specific codes used in the present study

I. BASIC SCIENCE AND TOOL DEVELOPMENT

1) *Basic science and other programs*: The project clearly focuses on causes, characteristics, developmental changes, and so on. This includes: (a) intervention research that does not focus on *clinical outcomes* typically and directly targeted by practitioners; for example, changes in a behavior not typically targeted in clinical settings or whose significance to date has yet to be established, or measures only used in research projects. (b) Infrastructure for basic science, or; (c) Programs to train researchers
2) *Development of tools and techniques:* In all cases, the project must address clinical outcomes as defined above. This includes: (a) developing assessment or treatment tools or platforms but without any actual testing of the tool with a clinical population; (b) developing training curricula or software associated with delivering services, or (c) work needed to prepare for a clinical trial.

II. PRE-IMPLEMENTATION IN SPECIALIZED SETTINGS

The project involves the use of specific and clearly defined clinical tools or techniques in *specialized settings* (e.g., a research clinic or a specialized hospital) on a *clinical population* (actually or potentially diagnosed with ASD or a related condition). These tools and techniques are considered to be eventually relevant to implementation. The setting is presumed to be specialized unless otherwise specified.

1) Pilot tools: The project pilots or seeks preliminary validation of a clinical tool or technique prior to implementation. This includes initial clinical trials, or projects exploring the moderating or mediating effect on a clinical target for a clinical technique. Validation of tools for parent training is coded here.
2) Specialized delivery: The project uses an validated clinical tool or technique in a specialized setting. The project may include extending the validation of a clinical tool or technique in a specialized setting. Parent training is always considered to involved implementation in community settings, and is coded below..

III. IMMEDIATE IMPLEMENTATION IN COMMUNITY SETTINGS

The project involves the use of a validated clinical tool or technique to address a clinical outcome on a clinical population, as defined above. *Community settings* include schools, the home, the workplace, and general hospital, an outpatient clinic, and so on. Any form of parent training is automatically included here.

1) Delivering services. This includes:
    a) Assessing delivery: The project assesses the use of a clinical tool or technique. This can include the first use of a new tool or technique in a community setting for the purpose of establishing its delivery. The project does not itself need to result in the delivery of a tool or technique; it can survey its delivery.
    b) Improving delivery: The project involves the delivery of a tool or technique, with the goal of improving its use. This includes the modification of a tool already in use in the community.

2) Closing gaps for *underserved populations* (e.g., those with less education or income, or from minority groups) in community settings. This includes:
    a) Assessing access: The project assesses access to a clinical tool or technique for an underserved population. The project does not itself need to result in the delivery of a tool or technique; it can survey gaps in its delivery.
    b) Improving access: The project seeks to improve access to a clinical tool or technique for an underserved population.
3) Building system capacity The project explicitly addresses the capacity to deliver a tool or technique through improved training, funding, policy, and programs. This includes:
    a) Assessing capacity: The project seeks to assess system capacity. This includes large scale studies seeking to establish the number of children diagnosed or treated.
    b) Building capacity: The project seeks to demonstrate how to increase system capacity

Figure 2: Decision Tree for the selection of project subcategories directly relevant to Screening and Diagnosis (Question 1) in Phase 1 (millions of dollars)

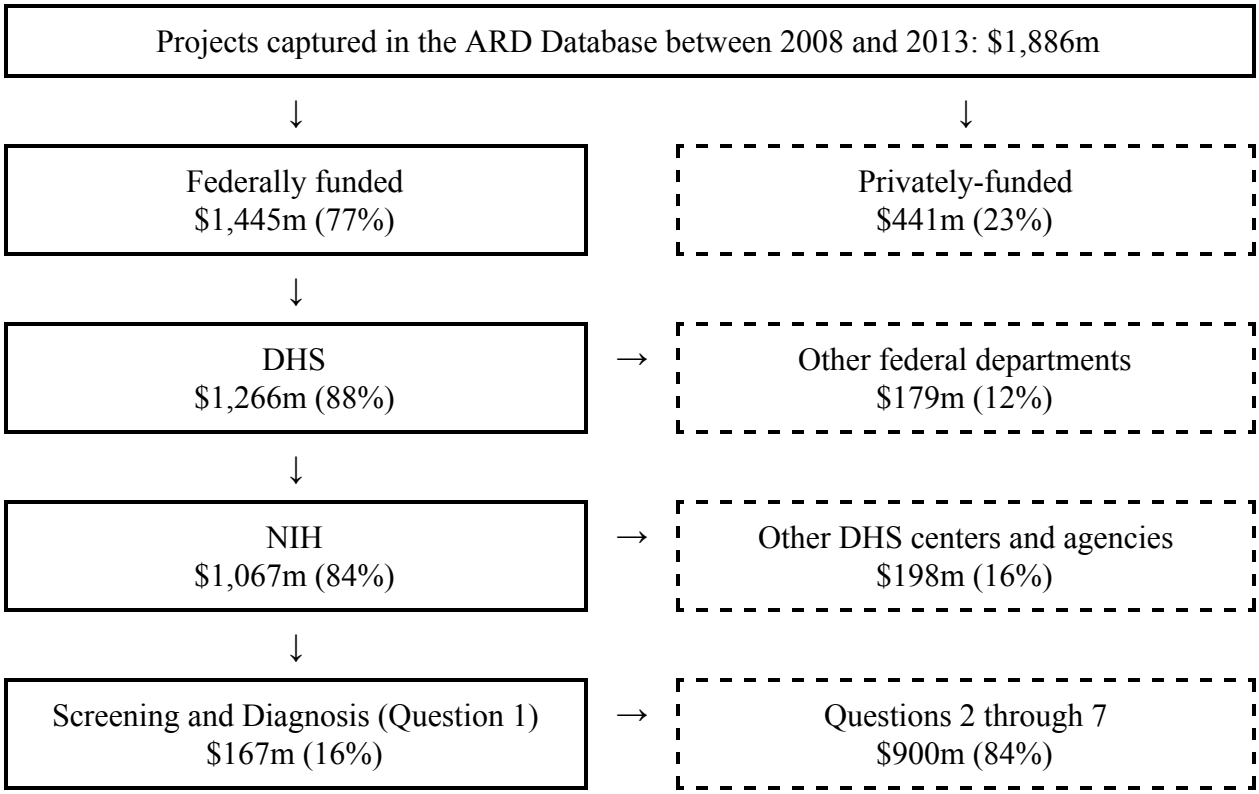

Figure 3: Funding of subcategories of projects focused on Screening and Diagnosis (Question 1) for 2008 to 2013, for NIH relative to other agencies

Subcategory	Funds allocated (% of total)	
	NIH	All Other Agencies
1.S.A. New diagnostic tool for diverse samples	$15.9m (18%)	$1.6m (3%)
1.S.B. Improved screening and diagnostic tool	$9.7m (6%)	$5m (11%)
1.S.C. Disparities in access to diagnosis	$0.5m (<1%)	$1.3m (3%)
1.S.D. Impact of early diagnosis of outcomes	$0	$0
1.S.E. Utility of genetic testing	$5.1m (3%)	$0
1.S.F. Ethical, legal, & social implications of research	$0.01m (<1%)	$0 (0%)
1.L.A. Behavioral and biological markers	$49.2m (29%)	$18.1m (38%)
1.L.B. Measures of behavioral / biological heterogeneity	$46.4m (28%)	$13.3m (28%)
1.L.C. Continuous dimensions	$10m (6%)	$1.8m (13%)
1.O. Other Questions	$30.5m (18%)	$6m (13%)
Total for question 1	$167.5m	$47.2m

Figure 4: Phase I Coding of IACC Subcategories for Screening and Diagnosis (Question 1), with levels of implementation

1. Basic Science: Utility of genetic testing (Q1.S.E); Ethical, legal, and social implications of research (Q1.S.F); Behavioral and biological markers (Q1.L.A); Measures of behavioral and/or biological heterogeneity (Q1.L.B); Continuous dimensions (Q1.L.C)

2. Pre-Implementation: a) Pilot Tools - New diagnostic tools for diverse samples (Q1.S.A), Improved screening and diagnostic tools (Q1.S.B)

3. Deliver Services: a) Assess Delivery - Impact of early diagnosis on outcomes (Q1.S.D)

4. Close Gaps: a) Assess Access - Disparities in access to diagnosis (Q1.S.C)

Not classified: Other (Q1.O)

Figure 5: Level of implementation of ARD subcategories of NIH projects focused on Screening and Diagnosis (Question 1) from 2008 to 2013

Level of Implementation	Funds allocated (% of total
I. PRE-IMPLEMENTATION	
1. Basic Science	$115.4m (69%)
2. Preparing for implementation	
a) Pilot tools	$25.6m (15%)
b) Specialized delivery	$0
II. IMPLEMENTATION	
3. Delivering services in community settings	
a). Assessing delivery	$0
b) Improving delivery	$0
4. Closing gaps for underserved populations	
a) Assessing access	$0.5m (<1%)
b). Improving access	$0
5. Building system capacity	
a) Assessing capacity	$0
b) Building capacity	$0
Other Questions	$26m (16%)
	$167.5m

Figure 6: Phase 2 coding of individual projects for Question 1