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ARTICLE

# Integration of stakeholder engagement from development to dissemination in genomic medicine research: Approaches and outcomes from the CSER Consortium

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ABSTRACT

**Purpose:** There is a critical need for genomic medicine research that reflects and benefits socioeconomically and ancestrally diverse populations. However, disparities in research populations persist, highlighting that traditional study designs and materials may be insufficient or inaccessible to all groups. New approaches can be gained through collaborations with patient/community stakeholders. Although some benefits of stakeholder engagement are recognized, routine incorporation into the design and implementation of genomics research has yet to be realized.

**Methods:** The National Institutes of Health–funded Clinical Sequencing Evidence-Generating Research (CSER) consortium required stakeholder engagement as a dedicated project component. Each CSER project planned and carried out stakeholder engagement activities with differing goals and expected outcomes. Examples were curated from each project to highlight engagement strategies and outcomes throughout the research lifecycle from development through dissemination.

**Results:** Projects tailored strategies to individual study needs, logistical constraints, and other challenges. Lessons learned include starting early with engagement efforts across project stakeholder groups and planned flexibility to enable adaptations throughout the project lifecycle.

**Conclusion:** Each CSER project used more than 1 approach to engage with relevant stakeholders, resulting in numerous adaptations and tremendous value added throughout the full research lifecycle. Incorporation of community stakeholder insight improves the outcomes and relevance of genomic medicine research.



## Introduction

Clinical genomics research has shown benefits through identifying diagnoses and enabling genome-guided care.[1,2] Despite these successes, the preponderance of research participants are from majority populations.[3,4] Inadequate representation of people of color and individuals with a lower socioeconomic status creates gaps in medical knowledge about effective approaches for screening, diagnosis, and treatment in these populations. These gaps impede equity in research benefits from clinical genomics research and contribute to disparities.

Numerous factors contribute to a lack of diversity in study populations, challenging researchers to examine and

*Correspondence and requests for materials should be addressed to Julianne M. O'Daniel, Department of Genetics, School of Medicine, University of North Carolina at Chapel Hill, Campus Box # 7264, Chapel Hill, NC 27599. *E-mail address:* jodaniel@med.unc.edu

A full list of authors and affiliations appears at the end of the paper.

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address potential barriers that may hinder access or willingness to participate.[5] Potential logistical barriers may include limited means of transportation to research appointments, competing demands for time, and the affordability of missing work.[6,7] Historically grounded mistrust of medical genetics and researchers[8,9] compounded with personal experiences of inequities in health systems, and poor communication with health care providers can further erode trust and contribute to skepticism of medical research. Moreover, research teams frequently lack the diversity of the communities they hope to enroll.

The Clinical Sequencing Evidence-Generating Research (CSER) consortium is addressing disparities in research participation via a concerted effort to recruit >60% of participants from underrepresented and underserved populations defined by each site on the basis of factors such as racial, ethnic, geographic, or insurance status data.[10] Thus, an additional directive at the programmatic funding-level for CSER was to engage with stakeholders to inform our diversity and inclusion efforts.[10] The clinical settings and aims for each of the CSER sites are described in Table 1.

## Community stakeholder engagement

Stakeholders are defined as representatives from the group(s) responsible for or affected by health and/or health care decisions informed by the research.[12,13] In clinical genomic research this includes diverse patients, parents, research participants, health care providers, payers, policy makers, advocacy groups, and community representatives.[14] Herein, we will focus primarily on engagement with community and patient/caregiver stakeholders. Such stakeholders can serve as cultural brokers that advocate for the needs and concerns of their community and build bridges between their communities and researchers.[15]

The engagement approaches and methods employed with community stakeholders are dependent on multiple factors. These include desired study outcomes, time commitments, relevance of issues, and experience of the stakeholders and researchers seeking to engage with them. Successful engagement necessitates researchers to be adaptable to change, to be willing to commit the time and resources required, to begin engagement early in the research process (preferably during idea generation), and to integrate feedback into research processes and outputs. This complexity and commitment may be daunting, but the benefits are essential. To encourage integration of stakeholder engagement, we highlight engagement strategies applied across the CSER projects framed by purposeful consideration of the approach and insights gained.

## Engagement across the research lifecycle

The key to engaged research is the application of strategies throughout the lifecycle of the research project. Figure 1 depicts the research lifecycle and potential opportunities for stakeholder engagement throughout the stages.

### Stage 1: Before award/planning

During the preaward period, study questions are defined, and hypotheses are formulated into specific aims and outcomes to propose for funding. At this early stage, having complete shared decision-making with stakeholders would align with the concepts of community-based participatory research,[16] which may be challenging in highly specialized fields and when there is limited time to respond to funding announcements. These challenges can be addressed through cultivating long-term relationships with stakeholders (eg, patients, advocates, and clinicians) such that readiness and capacity are already present in a standing advisory board. This affords ample opportunity to codevelop research questions, aims, and strategies with groups who are likely to be impacted by the research outcomes.

### Stage 2: After award/before enrollment

Once a proposal has received funding (or other initiation), preparation to conduct the study moves forward. Before enrollment, the study protocols, recruitment materials, consent processes, and educational materials will need to be finalized. This period offers robust opportunities to engage with stakeholders through a variety of mechanisms as described in the examples later.

### Stage 3: Ongoing enrollment

During this phase, participants who progressing through the study processes are actively enrolled. As challenges inevitably arise, there are unique opportunities to gain input from stakeholders to address emergent issues that could result in modifications to the study protocols and materials.

### Stage 4: Data analysis

The data analysis stage may be the most underused phase to incorporate stakeholder engagement in a genomic medicine research study. Some aspects may require specialized training and would not be practical to expect community stakeholders to perform. However, stakeholders may seek different questions from our data that can guide correlative analysis or examination for trends. In addition, stakeholder input into analysis can guide the framing of findings and development of key messages for future dissemination.

### Stage 5: Dissemination

Dissemination efforts may have diverse audiences, including the scientific community, clinicians, policy makers, funders, and organizational leaders as well as research participants and broader communities who may be impacted by current and future research efforts. Community stakeholder insight is essential to identify and prioritize key findings, guide lay language descriptions, and present opportunities to reach various community groups with broad and/or tailored messages. Their guidance can also shape professional community dissemination, including contributing to and coauthoring manuscripts[17–19] and copresenting at professional conferences.

**Table 1** CSER project descriptions

| Study Name[a] | Institution | Patient Enrollment Settings[b] | Target Enrollment Populations (Subpopulations)[b] | Key Outcomes |
|---|---|---|---|---|
| CHARM | Kaiser Permanente Northwest | Outpatient primary care clinics from 2 health systems:<br>Kaiser Permanente Northwest (Portland, Oregon) and Denver Health (a system of Federally Qualified Health Centers in Denver, Colorado) | Adults (18-49 years) at risk for hereditary cancer (racial/ethnic minority, low income, low health literacy, Medicaid/ Medicare or uninsured, Spanish speaking) | Assesses the utility of clinical exome sequencing and how it affects care in diverse populations of adults at risk for hereditary cancer syndromes. |
| ClinSeq | NIH/NHGRI[c] | NIH Clinical Center (Bethesda, Maryland) | Adults, no specific phenotype (African American, Afro-Caribbean, African) | Conducts genetic sequencing among healthy volunteers to study the impact of returning their individual genetic results and to build a resource for genotype-driven research. |
| KidsCanSeq | Baylor College of Medicine | Academic and nonacademic medical centers, outpatient clinics in Texas:<br>Texas Children's Hospital, MD Anderson Cancer Center (Houston); University of Texas Health Science Center at San Antonio, Children's Hospital of San Antonio (San Antonio); Cook Children's (Fort Worth); Vannie Cook Children's Clinic (McAllen) | Children with cancer and their parents (medically underserved, Hispanic/Latino, African American, Asian, Spanish speaking) | Studies the utility of genome-scale testing, compared with more targeted methods, in diverse pediatric patient populations with cancer and diverse health care settings in Texas. |
| NCGENES2 | University of North Carolina at Chapel Hill | Outpatient pediatric genetic and neurology clinics at academic medical centers; community hospital in North Carolina:<br>University of North Carolina Chapel Hill (Chapel Hill), Mission Health (Asheville), East Carolina University (Greenville) | Children (<16 years) and caregivers presenting as new patients with suspected genetic conditions (developmental disabilities, dysmorphology, neuromuscular disorders) (African American, Hispanic/Latino, Medicaid or uninsured) | Assesses the utility of clinical exome sequencing compared with standard of care testing in diverse pediatric populations presenting for initial genetic evaluation. Also assesses the impact of preclinic preparatory materials on measures of caregiver-provider engagement and care. |
| NYCKidSeq | Icahn School of Medicine at Mount Sinai & Montefiore Medical Center | Academic medical centers, private practice in New York City, New York:<br>The Mount Sinai Hospital, Mount Sinai Doctors Faculty Practice, Mount Sinai Kravis Children's Hospital, Mount Sinai West Hospital (Manhattan); Montefiore Medical Center, The Children's Hospital at Montefiore (Bronx) | Children (ages 0-21) with suspected neurologic, immunologic, and cardiac genetic conditions (African American, Hispanic/Latino, Medicaid, Spanish speaking) | Assesses the clinical and economic utility for use of genomic medicine for underserved children. Also assesses family understanding and satisfaction. |

*(continued)*

**Table 1** Continued

| Study Name[a] | Institution | Patient Enrollment Settings[b] | Target Enrollment Populations (Subpopulations)[b] | Key Outcomes |
|---|---|---|---|---|
| P$^3$EGS | UCSF | Academic medical center, outpatient clinics, neonatal intensive care unit in pediatric intensive care unit and community hospital in California: UCSF Benioff Children's Hospital Mission Bay, UCSF Fetal Treatment Center, Zuckerberg San Francisco General Hospital (San Francisco); UCSF Benioff Children's Hospital Oakland (Oakland); Fresno Community Health Center (Fresno) | Infants and children with severe developmental disorders, with or without congenital anomalies (pediatric); parents whose fetus has a structural anomaly (prenatal) (underserved by census tract, Medicaid, Asian, Hispanic/Latino, African American) | Assesses the utility of exome sequencing as a tool for diagnosing infants and children with serious developmental disorders. Also assesses providing genetic information to parents when a prenatal study reveals a fetus with a structural anomaly. |
| SouthSeq | HudsonAlpha Institute for Biotechnology | Academic medical center and community neonatal intensive care units, academic maternal fetal medicine outpatient clinics; Children's Hospital of New Orleans (New Orleans, Louisiana); University of Alabama at Birmingham (Birmingham, Alabama); University of Louisville (Louisville, Kentucky); University of Mississippi Medical Center, Woman's Hospital (Jackson, Mississippi) | Newborns with suspected genetic conditions (African American, underserved, rural) | Performs genome sequencing on newborns suspected to have genetic disorders. Assesses return of results mechanisms to expand access to genetic testing to diverse, especially historically underserved communities. |

Each CSER site is unique with different aims, processes, enrollment locations, and targeted enrollment populations.

*CSER*, Clinical Sequencing Evidence-Generating Research; *NHGRI*, The National Human Genome Research Institute; *NIH*, National Institute of Health; *UCSF*, University of California San Francisco.

[a]All projects have study materials (including consent forms, education materials, and surveys) available in both English and Spanish, some of which is publicly available at https://cser-consortium.org/cser-research-materials.

[b]Adapted with permission from Amendola et al[10] and Goddard et al.[11]

[c]ClinSeq completed enrollment at the start of the extramural studies and thus did not assess stakeholder engagement-related variables as in other CSER projects.

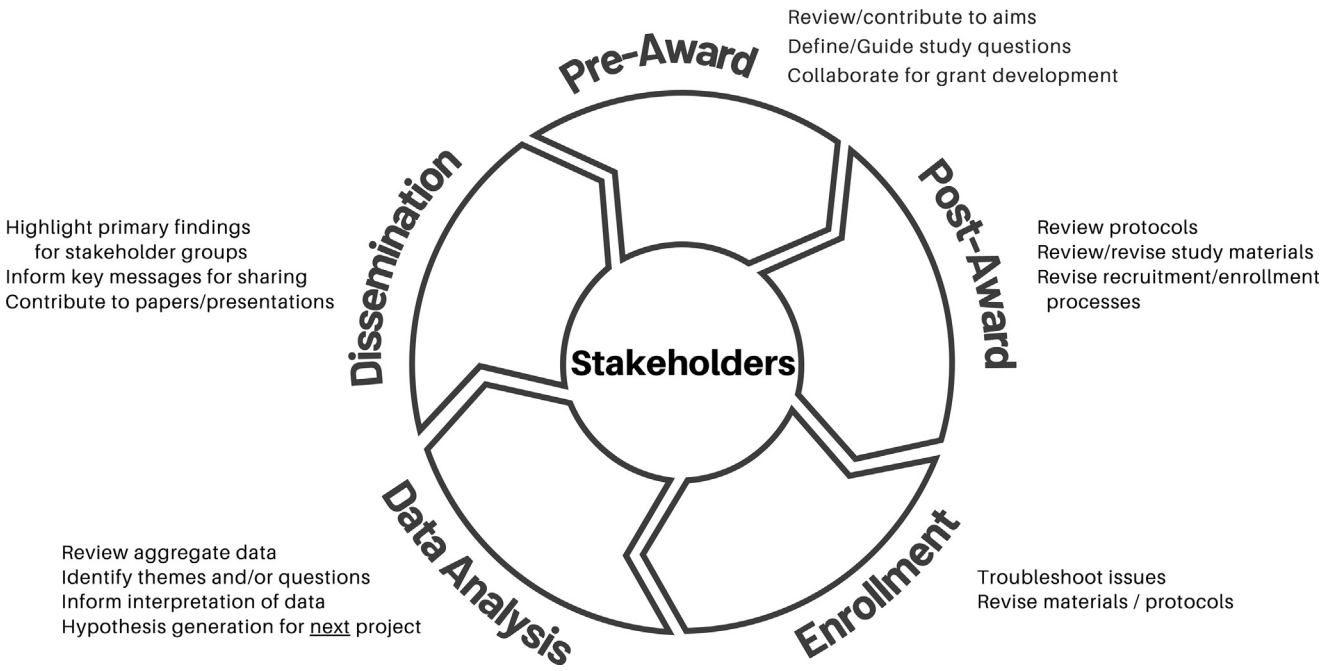


**Figure 1** **Stakeholder engagement** **throughout the research lifecycle enables continued value and insight.**

## Materials and Methods

Engagement methods are distinct from qualitative research because they are purposely not intended for the generation of new data. Activities typically include trust building with a target community; gaining insight into concerns relevant to the intended study population; development of culturally responsive research questions, materials, and protocols[20,21]; and troubleshooting research issues to identify alternate approaches that may be more responsive to the targeted participant population. Each CSER site is unique with different aims, processes, and targeted enrollment populations as described in Table 1.[10,11] Therefore, each project planned and carried out different stakeholder engagement activities with differing goals and expected outcomes (Figure 2). Approaches were selected from each of the active enrollment sites to demonstrate a variety of methods. The case examples describe how and why an approach was used and highlight some outcomes.

## Results

### Community engagement strategies

#### Advisory boards/action committees

Advisory boards and action committees ideally forge a partnership between researchers and various individuals who may have different roles in their community.[22] Consisting of community stakeholders, such as potential or previous research participants, advocates for patient groups and health conditions, social service professionals, and other types of community leaders (eg, faith leaders, attorneys, public service officials, community developers, and educators), they can provide valuable insights, iterative guidance, and recommendations as the research advances.

NYCKidSeq used a standing Genomics Stakeholder Board to enable partnership during the preaward/planning phase. The standing board is focused on implementing translational genomics in diverse New York City populations and includes patients, advocates, and clinicians, as well as researchers, funders, and entrepreneurs.[23] Beginning in the planning phase, NYCKidSeq research leadership met with the standing board to develop formative research questions.

Once awarded, the Genomics Stakeholder Board developed a project-specific action group including standing board members and newly recruited stakeholders to address the specific goals of the now funded project. In particular, this board helped to develop qualitative interviews and quantitative surveys for parents and clinicians, as well as Genomic Understanding, Information & Awareness, a low literacy, Spanish–English tool for genetic counselors to facilitate results disclosure.[24]

NYCKidSeq provided regular updates to their board about enrollment and retention by site and by recruiter. This enabled continual insight to help investigators address challenges and remain accountable for enrollment of various population groups. Based on feedback, the study conducted mock sessions to explore and address reasons potential participants were hesitant to enroll and added evening and weekend hours for study contacts. The close, ongoing relationship with the board was also instrumental to the team to rapidly and effectively pivot study contacts with participants to remote interactions due to COVID-19 clinical shutdowns.

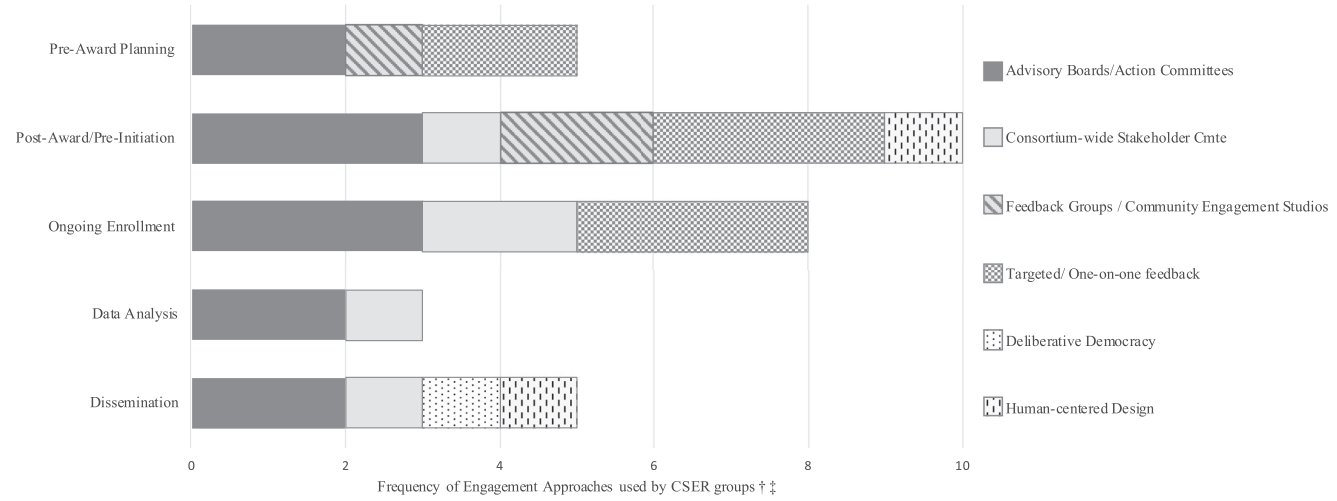


**Figure 2 The frequency of stakeholder engagement approaches used by CSER project groups across the research life cycle.** †Frequency refers to how many of the 6 active enrollment CSER sites indicated that they used that approach for activities in the particular stage. ‡The Consortium-wide Stakeholder Committee provided broad insight across the consortium and is therefore represented across each of the stages after award. In additional, they were used by KidsCanSeq for project-specific work during Ongoing Enrollment. CSER, Clinical Sequencing Evidence-Generating Research.

CHARM had 2 geographically distinct enrollment locations with different populations (Denver, Colorado and Portland, Oregon). They developed a Patient Advisory Committee (PAC) at each location during the postaward/pre-enrollment period to reflect the local population. PAC members included community members with low literacy skills, English and Spanish speakers, recruitment site staff, and study site staff engaged at the beginning of the study. Each PAC team held an initial in-person group meeting. For English-speaking members, subsequent meetings were also in person. Conflicts in schedules for Spanish-language PAC members prohibited group meetings; thus, all subsequent contacts with this group were conducted through targeted feedback via telephone. The PAC feedback influenced the development of recruitment and other participant-facing materials as well as study processes, such as consent and return of results.

NCGENES recruited and engaged a Community Consult Team (CCT) early in the postaward period to guide numerous elements of the project, including the development of a pre-clinic visit guide and question prompt list intended to impact caregiver-clinician engagement. Study participant families were randomized to receive/not receive these materials, and clinic visit transcripts from both arms were coded by research team members to explore question asking as one factor of engagement. The NCGENES coding team partnered with the CCT to analyze the coded data and explore the potential impact of the materials the CCT had developed. Their perspective provided insight into the complex nuance of information seeking, giving, and receiving that can occur during a specialist visit. CCT feedback was instrumental for informing data review and has been incorporated into discussions, ongoing analysis, and message framing for dissemination.

### Consortium-level community/patient stakeholder groups

Consortium-level community/patient stakeholder groups provide feedback and insight into the planning and implementation of research across a consortium. The CSER-wide community and patient stakeholder group consisted of 1 to 2 representatives recruited by each CSER project to represent their stakeholders. This group provided early perspective to the CSER consortium to inform research planning and partnership with participants. Key points included the following:)

- Keep information clear and simple. Do not overburden patients/caregivers who may already be overwhelmed.
- The distinction between research and clinical care may not be clear to participants.
- Participating in research requires building relationships and development of/maintaining mutual trust.
- Patients/caregivers want to work with researchers; treat them as partners, not menu items.
- Capacity building is not just for the community/patient stakeholders. Researchers also need to gain skills to appropriately engage.

The CSER-wide group provided a means for sites and consortium workgroups to engage with motivated, informed stakeholders through web-based conferencing. It also spread the potential challenge of recruiting and maintaining a group across multiple sites. This approach relied on a CSER project liaison to connect their stakeholder representative(s) to potential opportunities.

KidsCanSeq initially planned to use a standing Advisory Committee for the development and refinement of study materials; however, situational circumstances impeded that plan. The study team used the CSER-wide stakeholder advisory group for feedback about a specific process, namely to return variants of uncertain significance (VUS)

results via letter. Two virtual feedback groups (1 in English and 1 in Spanish) were held to review the types of result uncertainty that may arise and approaches to aid families' understanding of VUS. The summary points included preparing families for possible uncertainty, outlining what is known, and providing clear next steps. Providing resources and take-aways was encouraged. Using these points, KidsCanSeq developed an education card to include with results packets for participants who had a VUS identified.

A CSER consortium workgroup project aimed to describe a series of results discussion sessions collected from the ongoing CSER projects. Data analysis focused on themes regarding genetic counseling challenges mapped back to practice-based competencies. The work group project team engaged the CSER-wide stakeholder group through 2 virtual feedback discussions. Exemplar cases were discussed and possible counselor strategies were elicited and then paired with genetic counselor practiced-based competencies. Stakeholder input highlighted practical strategies the team had not considered, such as planning additional appointments to break-up discussion of complex topics and management.[25]

### Feedback groups

Feedback groups are small groups of stakeholders who represent specific experience or demographic characteristics for targeted feedback regarding a research project idea, recruitment plans or challenges, data collection methods, relevance and format of an intervention, and feasibility of the proposed research workflows. Guided by a facilitator and often using predetermined questions based on project-specific agendas, researchers can hold multiple feedback groups focused on the same topical agenda and/or different topical agendas at multiple points throughout the study. Community Engagement Studios (CESs) are a type of structured feedback group that typically uses an independent moderator not part of the research team and may be conducted in a different location with participants selected to have characteristics similar to the project enrollment needs.[26,27] CES enable studies to contract services through an experienced engagment team to facilitate stakeholder engagement. In both forms, research team members may choose to be present and listen to the discussion.

NCGENES used feedback groups before enrollment to gain insight into ways the research team could encourage enrollment by supporting future participant families on long clinical days. Caregiver participants from a previous genomics study were recruited to represent the target sociodemographic groups for the current study. These caregivers shared numerous practical suggestions based on their experience navigating specialty care with a complex-needs child and previous research participation. These included valet parking, substantial snacks, tablets with loaded content to entertain children, and participant "thank you" gifts for the child participant (eg, hats, t-shirts, and balls). Another outcome was insight into potential challenges that could arise for future participant families, including sickness, disruption in childcare, and transportation difficulties that could result in late or missed appointments. This emphasized the need for enrollment teams to have flexibility in scheduling research visits.

SouthSeq convened 2 CESs before enrollment. Parents of children who had previously been admitted to the neonatal intensive care unit at the Children's Hospital of Alabama were recruited through community outreach that included social media posting, flyers, and phone calls. Using CESs, the SouthSeq investigators received feedback on the Genome Gateway survey platform, the format and content of the genomic testing result letters, and the educational materials for parents aimed at improving knowledge about genomic testing. The CESs were conducted by trained moderators from Vanderbilt University, who were neutral to the project team. On the basis of the feedback obtained from parents, the research team changed the format of the genomic testing results template and revised educational materials.

### Targeted/one-on-one feedback

Targeted/one-on-one feedback seeks out individuals with specific knowledge, experience, or insights about a research topic,[28] who may be geographically distant or have other barriers to regular participation in groups.[29,30] As such, it may be particularly useful to engage traditionally underrepresented populations.

P$^3$EGS faced several challenges with recruiting and convening an advisory board that represented their study population with a high proportion of low-income and non-English–speaking participants. Barriers to attending scheduled group meetings included a lack of childcare support, inadequate transportation, and irregular work schedules. To better accommodate participating families and community stakeholders, the team pivoted to solicit one-on-one input on an as-needed basis. Feedback sessions yielded valuable information, including the need for and best way to provide incentives for completing study surveys as a way of respecting families' time.

KidsCanSeq used targeted feedback as a straightforward means to gain perspectives about their study process for return of results via mail for participants with no significant findings. The first 15 participants to receive results in this manner were contacted by phone and engaged for targeted feedback. Feedback led to a second effort to explore materials to help participants understand uncertainty stemming from genetics evaluation.

### Deliberative engagement/democracy

Deliberative engagement or democracy encompasses a range of approaches to seek public perspectives for the development of governing policies, such as those that guide health access or biomedical research.[31] Typically organized as a facilitated group or series of groups, this approach can engage diverse community stakeholders in deliberation with policy or situational experts with the aim of developing an informed consensus opinion or a set of recommendations to guide policy or practice development.[32,33]

SouthSeq has planning in progress to convene a multi-stakeholder group, including parents who with their

newborns participated in the research, members of the SouthSeq Community Advisory Board, clinicians, health system executives from the 5 medical centers participating in the project, and the investigators. The meeting will take place at the end of the project when study results are available for deliberation. A primary goal of the group is to produce informed stakeholder opinions, commentary, and feedback on the findings from SouthSeq to include in publications and other dissemination products. A secondary goal is to generate recommendations on the implementation of routine sequencing in neonatal intensive care units.

### Human-centered design

Human-centered design is a process that engages intended downstream users or beneficiaries in the design of a specific product or service.[34] It emphasizes multidisciplinary collaboration, creative brainstorming, and rapid prototyping and testing of solutions with the people who are most knowledgeable about a practice, service, or device that is in need of change.[35]

$P^3$EGS received supplemental funding to support the development of an innovative, human-centered design strategy for communicating aggregate study results to participants. Human-centered design is an approach to developing solutions that directly incorporate the values and priorities of intended beneficiaries.[36] The $P^3$EGS supplemental project is in progress and involves a multistage collaboration to (1) identify qualitative and quantitative study results that are of interest to participants; (2) develop simple, visually appealing materials in Spanish and English; and (3) disseminate results and assess their acceptability and relevance. The broader goals of the project are to increase public trust in scientific research and ensure that research participants share in the benefits of research by receiving information that is responsive to their needs and preferences.

## Discussion

For the benefits of genomic medicine to be accessible for all people, it is critical that clinical genetics research and outcomes be reflective of our diverse populations. Although the need for diversity is well supported, how to achieve that goal is intricately complex and potentially daunting for researchers. Patient and community stakeholders are a source of novel strategies and perspectives that are invaluable to enable the research shifts that are needed. However, routine integration of stakeholder feedback throughout a research project is not the norm. Often the term engagement is narrowly used to describe recruitment and retention efforts. Although research participants are certainly stakeholders in the research, we have purposely chosen to emphasize the added value of patient/community stakeholder engagement to inform the full research project. This means defining research questions, protocol planning, enrollment and troubleshooting, data analysis, and dissemination of key findings. This manuscript aims to provide working examples to encourage and empower researchers to incorporate stakeholder engagement into their research. Our intent is to demystify stakeholder engagement and shed light on potential obstacles, alternative approaches, and lessons learned. A brief comparison of the approaches highlighted are presented in Table 2.

### Identifying stakeholder groups

Research projects have numerous stakeholders. Although we have focused on community/patient stakeholder groups, it is important to recognize the existence and importance of engaging a broad range of groups who are involved in or impacted by clinical research. These include research team members, research participants (and caregivers), institutional review boards (IRBs), health care administrators, organizational leaders, payers, and funders. For example, some CSER sites faced barriers to implementation of their research because of difficulty in obtaining IRB approval—a challenge that has been documented by others[37] – likely exacerbated by the increasingly complex nature of genomic testing and research. CHARM researchers went on to analyze how interactions with IRB stakeholders shaped their project[18] and NYCKidSeq stakeholder members engaged their IRB to revise "standard" consent language to enhance understanding and clarity.

In addition, there may be a need to identify individuals and groups who can represent community/patient stakeholder perspectives that may be difficult to obtain directly. For example, some populations may be too emotionally vulnerable to be involved beyond participation as a research subject. This was faced by the research teams at $P^3$EGS, KidsCanSeq, and SouthSeq, whose target stakeholder group were caregivers of often very sick infants and children with potentially life-limiting conditions. Efforts to engage with vulnerable individuals requires sensitivity and consideration of the appropriateness of recruiting emotionally vulnerable stakeholders to participate in an advisory capacity. Engagement in an advisory capacity may be a positive experience for some participants and overwhelming for others. Therefore, researchers may need to consider alternative groups who can represent these key stakeholders. This can include caregivers who have previously experienced the situation or advocates for these families such as clinic social workers and community support leaders.

### Learning to be flexible

The importance of flexibility in approaching stakeholder engagement emerged as a key theme among all of the consortium sites. Community stakeholders are often busy, working adults who live in various locales. Some CSER projects amended plans to enable evening or weekend meetings or provide an option to call into meetings. Some sites found that rotating the location to different areas

**Table 2** Considerations for stakeholder engagement strategies: A comparison of the stakeholder engagement strategies used by the CSER project groups highlighting some benefits and challenges for each approach

| Approach | Benefits | Challenges |
|---|---|---|
| Advisory Committees | • Partnership between researchers and community<br>• Long-term relationship throughout the research project facilitates broader ideas and applications | • May not be representative of target population<br>• Requires significant time commitment from research team and stakeholders over course of the full project or as standing committee<br>• Requires transparency and shared decision-making |
| Targeted One-on-One Feedback | • Time and method can be tailored for convenience of the stakeholder<br>• Reduces constraints of transportation and childcare<br>• Increased privacy of discussion may enable deeper sharing<br>• Recruitment on an individual level can refine representativeness | • No group dynamic<br>• Limited number of stakeholders<br>• Feedback may be more individual compared with community/broader level<br>• Feedback may not be representative of target population |
| Feedback/Pilot Groups | • Group dynamics can introduce new and converging ideas<br>• Can enable perspective from larger numbers of stakeholders via separate groups<br>• Allows further exploration of responses through repeat meetings | • Discussion can be dominated by stronger personalities<br>• Need to coordinate space/time to meet needs of group (eg, evenings, weekends)<br>• Need to consider other supports to enable participation (eg, travel, food, childcare) |
| Community Engagement Studios | • Facilitated by a neutral moderator<br>• Enables teams to tap into readily available expertise and processes | • Representative populations recruited by moderator may be different than target study population<br>• Consulting fees for facilitators may be higher than internal teams |
| Consortium Stakeholder Group | • Representation across the consortium projects<br>• Enables broader discussion on shared themes<br>• Increases diversity and/or generalizability of perspectives<br>• Interaction with/influence on consortium leadership and funders | • Need to consider scheduling, communications and budgeting logistics across a broad geographic area (eg, multiple time zones)<br>• Issues and/or feedback may not be applicable to a specific project<br>• Consortium-level issues may not be as relevant to the local project stakeholder leading to less feedback |
| Deliberative Democracy | • Includes community voice from the population studied<br>• Includes relevant subject matter experts on the target issue<br>• Aims to propose policy or opinion about a defined issue through consensus building<br>• Resulting policy/proposal reflects the informed opinion of the stakeholders | • Significant time commitment from research team and stakeholders over a defined period of time (eg, multi-day event)<br>• May involve recruiting large numbers of stakeholders representing multiple perspectives<br>• Can require a team of facilitators to coordinate<br>• Often includes significant capacity building through informational presentations to enable informed deliberation |
| Human-Centered Design | • Iterative process that involves direct collaboration with stakeholders<br>• Participatory design can lead to more relevant, meaningful, and useful outcomes | • Significant time commitment from research team and stakeholders over multiple interactions<br>• Stakeholders may not be representative of overall participant population |

*CSER*, Clinical Sequencing Evidence-Generating Research.

ensured the travel burden was reduced and equally shared. Sites also worked to build relationships in virtual group meetings held through web-based meeting platforms.

Research teams also tailored engagement strategies to meet the study needs and facilitate stakeholder participation (Figure 2). All CSER sites endeavored to engage stakeholders from underserved and underrepresented populations. To successfully do this, research teams could not use a one-size-fits-all approach and needed to adjust planned methods. For example, the P$^3$EGS study initially planned to convene a traditional community advisory board but found that convening such a group in person presented challenges for families with unpredictable work hours and caring for a child with special needs. Instead, the P$^3$EGS team pivoted to use one-on-one discussions with stakeholders, which also facilitated inclusion of participants who did not speak English.[38]

The concept of flexibility also applies to the broader research timeline. Research timelines are critical-period. Competing research priorities and the typical hiccups common to clinical research can diminish the resources available and value placed on stakeholder engagement activities. Emphasis in translational genomics research can be hyperfocused on enrollment, retention/attrition, and numbers of laboratory tests completed. The additional time and budget required to establish robust stakeholder relationships can be seen as overly burdensome especially if it means delaying enrollment or other key metrics required in quarterly reports. To be successful, research team leadership needs to value stakeholders as integral to the research timeline, recognize how stakeholder guidance can help meet the deliverables (ie, recruitment), and plan for the appropriate time and flexibility to ensure stakeholder insight throughout the project.

### More than just talk—Integrate

Stakeholder engagement is not a one-way street in which researchers explain what they are doing to their advisory group. Researchers must be willing to amend protocols, reconsider data questions, and actively partner on message framing and dissemination. CSER projects incorporated stakeholder feedback into protocols and materials. Sites that engaged advisory boards continued to incorporate that feedback throughout their studies. Actually incorporating stakeholder feedback and sharing changes made with the broader research community demonstrate authenticity and show stakeholders their input is critical and meaningful.

## Conclusion

Each CSER project used more than one approach to engage with relevant stakeholders, resulting in numerous adaptations and tremendous value-added throughout the full research lifecycle. Early and continual engagement offers the opportunity for research questions, aims, and strategies to be influenced by the groups who are likely to be impacted by the research outcomes. Mid-project engagement affords the integration of stakeholder insight into study logistics such as participant-facing materials (eg, consents, recruitment, etc.), enrollment protocols, and troubleshooting for protocol or enrollment-based challenges that inevitably arise. Notably, each project successfully met or exceeded their goals to enroll >60% research participants from underrepresented populations. Continued engagement throughout data analysis and dissemination enables stakeholder perspectives to influence the analysis of findings, highlight key messages, and inform dissemination to diverse audiences most likely to be impacted by the research.

Incorporation of community stakeholder insight throughout research projects can improve medical genetic research and outcomes. For our research to benefit all populations, it is imperative that stakeholder engagement efforts be recognized, valued, and supported as integral, not merely supplementary, to medical genetics research.

## Data Availability

All data supporting the engagement methods described and outcomes gleaned are available from the authors on request. Most of these data are contained in project publications, which have been referenced when available.

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## Clinical Sequencing Evidence-Generating Research diversity, equity, and inclusion statement

In the Clinical Sequencing Evidence-Generating Research consortium, we aim to improve the use of genetic information in medicine and reduce barriers to genetic services among underserved groups. Our research seeks to better understand connections between genes, other drivers of health and disease, and health outcomes. We have worked with study participants and community partners to help make our research more inclusive. We still have much more work to do to ensure that our findings are applied in fair and just ways. We also acknowledge the need for more diversity among our own researchers. As we publish the results of Clinical Sequencing Evidence-Generating Research, we

commit to carrying efforts forward to make sure people of all backgrounds benefit from genomic research and medicine.

## Author Information

J.M.O., L.M., L.R.D., S.A., S.J.K., G.J., S.R., C.R.H., and E.B.M. significantly contributed to the conceptualization of this manuscript. J.M.O., S.A., L.R.D., S.R., S.J.K., K.P.A., M.I.D., C.R.H., N.M.L., C.K.M., K.F.M., and C.S. assisted in curating project-specific data and methods cases from each site. J.M.O., S.A., L.R.D., S.R., S.J.K., L.M., C.R.H., and G.J. contributed to the initial drafts. J.M.O., S.A., L.R.D., S.R., S.J.K., L.M., G.B., K.P.A., M.I.D., C.R.H., G.J., G.L., N.M.L., C.K.M., K.F.M., M.A.R., M.R., C.S., E.B.M. reviewed and edited the final manuscript.

## Ethics Declaration

This manuscript presents methods and approaches. Each project described obtained ethics review through their institutional review boards as appropriate.

## Conflict of Interest

The authors declare no conflict of interest.

## Authors

Julianne M. O'Daniel[1,*], Sara Ackerman[2], Lauren R. Desrosiers[3], Shannon Rego[4], Sara J. Knight[5], Lonna Mollison[1], Grace Byfield[1], Katherine P. Anderson[6], Maria I. Danila[7], Carol R. Horowitz[8], Galen Joseph[9], Grace Lamoure[10], Nangel M. Lindberg[11], Carmit K. McMullen[11], Kathleen F. Mittendorf[12], Michelle A. Ramos[8], Mimsie Robinson[13], Catherine Sillari[10], Ebony B. Madden[10]; On behalf of the CSER Stakeholder and Engagement Work Group

## Affiliations

[1]Department of Genetics, School of Medicine, University of North Carolina at Chapel Hill, Chapel Hill, NC; [2]Department of Social & Behavioral Sciences, School of Nursing, University of California San Francisco, San Francisco, CA; [3]Department of Pediatrics, Baylor College of Medicine, Texas Children's Cancer Center, Texas Children's Hospital, Houston, TX; [4]Institute for Human Genetics, University of California San Francisco, San Francisco, CA; [5]Division of Epidemiology, Department of Internal Medicine, University of Utah School of Medicine, Salt Lake City, UT; [6]Denver Health Ambulatory Care Services, Denver, CO; [7]Division of Clinical Immunology and Rheumatology, Department of Medicine, Heersink School of Medicine, The University of Alabama at Birmingham, Birmingham, AL; [8]Institute for Health Equity Research, Icahn School of Medicine at Mount Sinai, New York, NY; [9]Department of Humanities and Social Sciences, University of California San Francisco, San Francisco, CA; [10]National Human Genome Research Institute, National Institutes of Health, Bethesda, MD; [11]Center for Health Research Kaiser Permanente Northwest, Portland, OR; [12]Vanderbilt Ingram Cancer Center, Vanderbilt University Medical Center, Nashville, TN; [13]Bethel Gospel Assembly, New York, NY

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