Participant-centric initiatives: Tools to facilitate engagement in research

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A B S T R A C T

Clinical genomic research faces increasing challenges in establishing participant privacy and consent processes that facilitate meaningful choice and communication capacity for longitudinal and secondary research uses. There are an evolving range of participant-centric initiatives that combine web-based informatics tools with new models of engagement and research collaboration. These emerging initiatives may become valuable approaches to support large-scale and longitudinal research studies. We highlight and discuss four types of emerging initiatives for engaging and sustaining participation in research.

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1. Introduction

Despite increasing capabilities in technology and analytic perspectives, translational researchers still face familiar challenges in generating successful trials and studies (Collins, 2011). Both in the US and internationally, slow recruitment and limited retention of research participants can hinder the research process (Sung et al., 2003; Center for information and study on clinical research participation (CISCRP), 2012; Gottweis et al., 2011). Participation and engagement are further limited within some marginalized socioeconomic or cultural groups (Gottweis et al., 2011; Bowen and Penchaszadeh, 2008; Wynia and Gamble, 2006; James et al., 2008; Bussey-Jones et al., 2010). In parallel, human subjects’ research regulations in the U.S. and E.U. pose ethical and public relations challenges (Silverman et al., 2001; Fullerton and Lee, 2011), with many researchers and institutions preferring to use de-identified datasets to limit responsibilities and liabilities. Against these common research challenges are evolving new modes of data sharing and collaboration, including research networks and increasingly coordinated access to large-scale resources such as biobanks or clinical data warehouses. These innovations also increase the complexity of coordination, accountability, preference management, and researcher-participant communication (Fullerton et al., 2010; Heeney et al., 2011; Kaye et al., 2009; Mascalzoni et al., 2008; McGuire et al., 2011; Ludman et al., 2010; Harmon, 2010). The ability to leverage communications technologies such as social media may ameliorate some of the traditional roadblocks to broad patient participation in health care (Trinidad et al., 2011), and similar approaches provide new models for engaging participants in the research process and facilitating researcher-participant collaboration. In clinical settings, decision aids have been evolving to support patients’ self-education and decision support as components of their participation in therapeutic paths, and these methods can now also play a part in research (Swan, 2009). As these technologies and challenges intersect, a range of public/private participant-centric initiatives illustrate how information tools can expedite translational research. We highlight four emerging types of initiatives that illustrate evolving approaches of participant engagement and the use of informatics-based tools to expedite translational research.

2. What are patient centric-initiatives?

Participant-centric initiatives (PCI) are tools, programs, and projects that empower participants to engage in the research process and, in many cases, can differentiate between a range of diverse preferences and needs. Although current U.S. and E.U. human subjects’ regulations permit secondary research on de-identified data and biosamples without further participant contact or consent (Fullerton and Lee, 2011), cautionary tales demonstrate that people feel that they are marginalized if they are excluded from the research process (Ludman et al., 2010; Harmon, 2010). For example, some research participants are not concerned about what happens to biosamples that have been collected from them for research, yet many participants have concerns about their lack of involvement in data sharing for secondary research use (Trinidad et al., 2011, 2010; Brase, 1998). Diverse preferences need dynamic tools to manage them, and one-size fits all approaches such as waivers of consent or “broad consent” are increasingly under significant critique (Simon et al., 2011; Sheehan, 2011; Hansson et al.,...
2.1. Finding the engaged participant through intermediation: participant–researcher ‘matchmaking’ tools

A range of potential factors may affect consideration of participation in research, including prior participation in research and existing relationships with researchers, involvement of trusted leaders, and trust in the organization (Gottweis et al., 2011; Bowen and Penchasazdeh, 2008; Wynia and Gamble, 2006; James et al., 2008; Bussey-Jones et al., 2010). Although a majority of US residents (77%) say that they would consider becoming involved in a research trial, only 10% of those eligible to participate do so (Center for information and study on clinical research participation (CISCRP), 2012). Many are not aware of research opportunities (unpublished results, NWABR 2012). European residents vary considerably in consideration of contribution of data or samples to a biorepository for research purposes, with “93% of Icelanders and 82% of Norwegians could imagine providing information to a biobank, but only 25% of Latvians or 35% of Austrians are likely to do so” (Gottweis et al., 2011). Some common barriers to recruitment include lack of participant incentives, privacy concerns, complex and confusing consent forms, differences in cultural norms, and lack of public knowledge about the potential for participation. One possible solution is more active facilitation of connections between willing volunteers and researchers, and enhanced communication structures that support these potential relationships. Matchmaking tools do so by acting as intermediators that draw together researchers and participants. For example, participant-driven clinical registries, such as ResearchMatch.org, TrialX.com, and EmergingMed.com enable willing volunteers to enter public knowledge about the potential for participation. One possible solution is more active facilitation of connections between willing volunteers and researchers, and enhanced communication structures that support these potential relationships. Matchmaking tools do so by acting as intermediators that draw together researchers and participants. For example, participant-driven clinical registries, such as ResearchMatch.org, TrialX.com, and EmergingMed.com enable willing volunteers to enter

Figure 1. Example of modes of engagement and communication for patient-centric initiatives.
and researchers. Continued technological advancement and wide-scale adoption by researchers and volunteers alike are contributing to richer data resources, and are demonstrating the potential of such matchmaking tools for overcoming recruitment bottlenecks in limited domains, though a comprehensive evaluation of the effectiveness of these tools across a range of demographic and disease types is still nascent.

2.2. Engagement through direct participant benefits: ‘Direct-to-consumer’ tools

Direct-to-consumer (DTC) tools offer individuals highly personalized information as a proposed direct benefit of participation. For example, established web-based DTC genetic testing companies, such as 23andMe.com or deCODEme.com seek to empower and promote identity perception for individuals by supplying them with personal genotype and phenotype information that may inform their personal health decisions (Nordgren, 2012). The social interactions between DTC companies and their customers are not without controversy, as the promise of this form of consumer-focused health decision making has to date managed to avoid either the predicted dire consequences of a excessively worried public, or the proposed swell of empowered and activated patients (Nordgren, 2012; Cecile et al., 2010). What the DTC companies have enabled beyond personalized information is the increased ability to facilitate participation in research. The research arm of 23andMe, 23andWe, notifies members of research opportunities enabled through more personalized matching of studies than ResearchMatch through an extensive set of structured surveys about their phenotypic traits. By connecting those self-reports to corresponding genotypes, 23andWe can produce publishable study results, and within days can verify other published results (Tung et al., 2011; Wicks et al., 2011). To help drive their research arm, 23andMe created condition-specific member communities (e.g., Parkinson’s, sarcoma, pregnant women). By joining, members receive free genetic testing and involvement in the community forum. For example, the “Roots into the future” community is an up-and-coming community that offers 10,000 African Americans free genetic testing in return for completing surveys. 23andWe’s model leverages individual’s willingness to participate in research and rewards their participation with information people value. Given the amount of genetic information that 23andMe can gather through this approach to participant engagement, it is reasonable to assume that they have the potential to produce many more substantial findings.

Like 23andMe, the health social network PatientsLikeMe.com started as an organization that provided a service to people curious about their health and frustrated with the slow progress of traditional research. PatientsLikeMe.com has a substantial research arm and has great potential to conduct studies faster and cheaper than existing models (Tung et al., 2011). For example, analysis of data reported by people with amyotrophic lateral sclerosis (ALS) who experimented with lithium (Eolgin, 2010). Members of Genomera.com can develop their own study designs (i.e., study creator), participate in the study themselves, and post procedures for other community members to participate in their study. Study participants then send their results to the study creator for analysis. The openness of this community enables participants to be recruited and studies to be completed much quicker than the traditional research process.

2.3. Participant control through choices: tools for ‘dynamic negotiation’ between researchers and participants

DTC companies seek to offer something that many research institutions are unable to provide – a user-friendly interface that gives users of a range of literacy and education levels descriptive, personalized and easily accessible information. However, at present most DTC users pay for the service, and in doing so, fuel research efforts of the parent companies offering these services. Although most researchers cannot individually offer the same usability and customer support amenities as 23andMe.com, they are increasingly able to offer participants greater levels of choice and control, as well as manage a diverse range of participant preferences. The company Private Access (privateaccess.info) is one such platform that seeks to provide participants with substantial control over the uses of their data in research (Terry and Terry, 2011). The platform allows researchers to search for potential study participants with great specificity because participants who use Private Access enter their entire personal health records, and have control over both individual portions of their personal health records as well as the scope of researchers and research groups that the data is visible to. Researchers can use Private Access to recruit participants, access this private data by being granted “private access” by the participants, and use the data in the records of recruited participants in applicable research projects.

Private Access uses this dynamic negotiation approach to moderate patient involvement in research repositories. The traditional practice of entering data into repositories often requires participants’ broad consent to authorize secondary use of their anonymized or de-identified data. Although it may be impossible for participants to fully know all potential future studies in which their data could be used, refusal to agree to such broad use thus renders secondary usage of their data an all or nothing decision (Fullerton and Lee, 2011). Private Access arguably overcomes this bottleneck by permitting individual control over privacy and data sharing, much like social sharing tools like Facebook.com. Some participants may choose to set liberal privacy settings, permitting any researcher access to their data in full for screening and eligibility purposes, while others may request to be contacted about study eligibility after giving access permission on a case-by-case basis. By giving participants this dynamic control, Private Access supports broad consent, if that is what an individual prefers, but it also gives participants the opportunity to choose the access level with which they are most comfortable over time. It should be noted that much like Facebook and other social-networking sites, the definition of what is personally and culturally appropriate to share is an evolving discussion and one that is likely to vary from person to person. Tools such as Private Access, or the open-source Indivo (indiviohealth.org), can be tailored to provide culturally specific information or utilize trusted community leaders as “guides” through the decisions involved in selecting preferences. Data is emerging now regarding how tools such as these facilitate participant participation.

2.4. Public engagement through citizen science: direct participant involvement tools

Increased public awareness of large volunteer registries, such as the Love/Avon Army of women (armyofwomen.org) or Inspire 2 Live (inspire2live.com) can be considered a minimal form of public engagement in the research process. Towards the other end of the spectrum lies Genomera.com, a company that seeks to empower and engage the public in research by giving them the opportunity to be ‘citizen scientists’ who can design, conduct, and analyze their own studies (Eolgin, 2010). Members of Genomera.com can develop their own study designs (i.e., study creator), participate in the study themselves, and post procedures for other community members to participate in their study. Study participants then send their results to the study creator for analysis. The openness of this community enables participants to be recruited and studies to be completed much quicker than the traditional research process.

Given the established conventions for peer review and controlled research, the likelihood of the citizen scientists at Genomera.com having their results published in scientific journals is presently slim.
However, with widespread adoption of this approach, validation of methods, the possibility of finding significant results, or creating methods that can be replicated in other studies is possible. Furthermore, such citizen science approaches are challenging existing paradigms about acceptability of standards in research, finding their own distribution channels outside of traditional dissemination strategies (CES4Health.info, 2012). Efforts, such as Genomera.com, demonstrate the desire for people to play a larger role in research. Whether through educating participants about studies they participate in, returning study results (Beskow and Smolek, 2009), or answering participants’ questions, participants can become more involved (Gust and Seifer, 2011). Involving participants could increase the likelihood of future participation, adhere to study protocols, or sharing positive research experiences with others.

3. Conclusion

There are widespread changes occurring internationally in health care, and all face common challenges of effective leverage of information technology, the need for accurate clinical and health data, and the need for privacy protections (Collins, 2011; Meslin and Cho, 2010; U.K. E-Health Records Failure Makes U.S. Plan Shine, 2011). The research enterprise can utilize similar developments in order to keep up with changing socio-cultural context that requires more engaged research participation to be successful. If decisions are made and practices are built without due diligence to public opinion, then the string of inefficiencies and ethical questions will unnecessarily grow. We have observed resources destroyed due to of lack of appropriate public engagement (Root, 2010). We should look not only to public opinion, but also to the empowered public by facilitating their engagement in the research process as key stakeholders. Recruiting participants, protecting their privacy, and ensuring informed consent should not be viewed as burdensome bottlenecks, but rather as opportunities to engage, inform, and benefit the ultimate end-user of all research, the public.

Using PCI in research is one approach to overcome these challenges by leveraging new communication and facilitation modes increasingly available through the on-line economy. From the informatics-based initiatives we have presented, to the great successes of participant-centered organizations like Love’s Army of Women and Genetic Alliance, to participant-driven social networking sites, the possibilities of an empowered public are starting to be realized. Data are needed in a variety of settings to test whether and how PCI can facilitate and sustain research participation across populations, particularly those with less access to web-based technologies and who may benefit the most. If we keep lessons learned from community-based research, minority recruitment, decision support, and other innovations, we can meet the goal of an active and invested community of research participants. With the current initiatives already in development and use, we have crossed an important threshold of feasibility testing and can now move into efficacy and effectiveness studies. Research groups and funders can begin to make the choice to utilize these tools and study the process along the way.

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