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Perspective of Patients and Stakeholders as Members of a Research Team

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Abstract

Systemic sclerosis (SSc) is a rare orphan disease, characterized by skin thickening, vascular insufficiency, and fibrosis of internal organs. SSc affects about 100,000 people in the United States. This study explored perceived benefits and challenges of patient partners and stakeholders, who were team members on a project to revise and test a self-management program (Taking Charge of Systemic Sclerosis). Five patient partners, 1 stakeholder from the Scleroderma Foundation and 1 stakeholder from a state chapter of the Scleroderma Foundation were interviewed. Conversations were audio recorded and transcribed verbatim and analyzed. Four themes emerged from the analysis with corresponding subthemes: contributions to study, benefits of involvement, challenges, and project leadership. The themes and subthemes were generally similar to those expressed in other studies. However, additional benefits from engagement were identified: acceptance, increased knowledge of SSc, and helping others. Participants reported feeling supported and valued as members of the team and that their opinions mattered which is in contrast with findings from other studies.

Keywords

systemic sclerosis, patient engagement, patient education, qualitative

Introduction

Engagement of patients and/or stakeholders in research is recognized as a standard to improve the relevance of transferring research findings into practice (1-6). Engagement includes many roles (eg, consultants, co-investigators, research subjects) (5,7) and occurs in separate phases and stages of research (eg, setting priorities; identifying topics, questions, interventions; interpreting findings; and disseminating findings) (6-8).

Benefits of engagement from the perspective of the patient and/or stakeholder have been identified as being recognized as knowledgeable experts, identifying and prioritizing research questions, identifying outcomes important to patients, recruiting participants, and developing user-friendly recruitment materials (1,6-10). Challenges included tensions between scientific rigor of a study and adhering to a protocol versus stakeholder views of research and less rigid adherence to protocols, researchers' tokenistic views of patient stakeholders, and time and costs associated with attending meetings. Suggestions to alleviate challenges were identified and adopted in the present study (1,6,8,11).

Patient and stakeholder engagement is especially crucial when the population of interest has a rare orphan condition such as systemic sclerosis (SSc). SSc affects about 100,000 people in the United States (12) and is characterized by skin thickening, vascular insufficiency, and fibrosis of internal organs (13). Because of the rarity, many people do not have access to education programs or support groups. To address the lack of programs, a self-management program (Taking Charge of Systemic Sclerosis, TOSS) was developed with input from people with SSc (14,15). As new pharmacological treatments and changes in recommendations for laboratory and diagnostic tests became available, the developers undertook a major revision of TOSS (16). This revision afforded opportunities to systematically engage patient partners and stakeholders, in all phases of research, to ensure that the content and product were valuable and useful (16).

Previous studies on the value of engagement included participants who had been engaged in different studies, thus, working with different research teams. This study is unique

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Table 1. Semi-structured Interview Questions.

- As a patient partner and/or representative of a stakeholder organization, what do you feel was your most important contribution to the study?
- What were positive / benefits or negative aspects to you from being a member of the research team?
- What did you enjoy most about being a member of the research team? What did you enjoy least?
- How do you think the study benefited from having you as a patient partner or representative from a stakeholder organization on the team?
- Are there any other ways you would have liked to be involved as a member of the research team? What other ways?
- Based on your experiences with this project, what do you think are the best ways to engage patient partners and/or representatives from stakeholder organizations as research team members?
- What do you think the next steps should be with the TOSS internet program? How do you think we should disseminate it to the scleroderma community?
- Do you think translation to another language is important? What language?

as we explored perceived benefits and challenges of patient partners and stakeholders, who were team members on the same PCORI funded study.

Methods

Within a month of study completion, all patient partners and stakeholders who were members of the research team for a PCORI funded study, from grant submission (2014) to study completion (2017), were contacted by JLP by email and invited to participate in the study. We chose to interview participants at the end of the study in order to obtain global perspectives over the lifetime of the project. Participants included 5 patient partners, 1 stakeholder from the Scleroderma Foundation, and 1 stakeholder from a state chapter of the Scleroderma Foundation. One additional patient partner was invited but could not participate due to other obligations and time commitments. Participants provided written informed consent, and the study received approval from the university human protection office.

Participants were interviewed individually on the telephone by SLN, an experienced interviewer and qualitative researcher, who was also a member of the research team. Interviews took about 60-90 min and were audio recorded. Interview questions are in Table 1. Recordings were transcribed and all identifying information removed from the transcripts during the verification process.

The de-identified transcripts were imported into NVivo 11 (QSR International, Melbourne, Australia) and analyzed for emergent themes through 2 stages of content analysis: (1) coding by one researcher to create an initial coding structure grounded in the discussion topics described above and (2) a process to verify and expand this coding to include sorting of discussions into emergent themes and categories. A second team member, JLP, conducted an independent reading of transcripts and edited the initial coding structure. Any discrepancies were resolved by discussion. Guidelines from the Consolidated Criteria for Reporting Qualitative Research (COREQ) were followed to ensure research quality (see Supplemental Table 1) (17).

Results

Mean age of participants was 55.14 years (SD = 13.4), mean education level was 18.3 years, 71% were women, 71% were white, 71% were married, and 57% worked full time. Of the participants with SSc, 57% had diffuse SSc and the mean time since diagnosis was 13.8 years (SD = 11.1).

Analyses revealed 4 inter-related themes and 15 subthemes of patient experience. These are displayed in Table 2, along with quotes that illuminate the voices of the patient partners and stakeholders.

The overarching subtheme, under "**Contributions to the Project**" was *patients having SSc*. Patient partners felt they brought a unique perspective of living with SSc and the needs of that group and their families. Even though the stakeholders did not have SSc, they were familiar with the SSc community and recognized the contributions the patient partners made. One voiced "when you pull in patients, you are going to have the best possible outcomes." In addition, participants mentioned *uncertainty and fear*. They felt the TOSS website should become the first place newly diagnosed patients should come to for credible information to help alleviate some of the concerns. One said "it [TOSS] actually have me more enthusiasm and more hope." The subtheme, *active participation in study protocols*, pertains to the contributions patients partners and stakeholders made in creating materials for TOSS, facilitating focus groups, and selecting instruments to be used as outcome measures. Our patient partners and stakeholders created sections of modules for TOSS, audio testimonials, and resource materials. One participant said "I wanted to make it [TOSS] very meaningful so when they went on the website it was something they could take away and make it applicable to their everyday life." Both males mentioned gender as adding value as they provided input on what was needed from the male perspective as a lower percentage of males have SSc. Our Spanish speakers recognized a need for a Spanish translation.

The theme, "***Benefits of Involvement***" included several subthemes, one of which was *engagement with research process*. Most participants, both patient partners and stakeholders, had only been involved with research as participants. For these individuals, observing the evolution of the study

Table 2. Themes, Subthemes, and Representative Quotes From Interviews.

Themes	Subthemes	Illustrative quotes
Contribution to study	• As patient with scleroderma knowledge • Reduce fear	*Because we all are so familiar with scleroderma because we're involved in it almost on a daily basis… So, that's the biggest thing I think I brought. Just for us to be sensitive to the new uneducated scleroderma patients and family members of scleroderma patients.* *I think just contributing from a patient's perspective and also with the background that I have. I think patient involvement is critical. As researchers, we can just get sort of off base sometimes. So, I think the patients bring us back to what we should really be focusing on here.* *I was hoping that my voice was heard when I said, let's not scare people. Because most of the people who are going to come to us are first timers or novices and scared. Scared as heck just because they cannot say the word scleroderma.*
	• Active participation in study protocols ○ Create useful materials for TOSS ○ Focus group facilitation ○ Instrument selection	*I think that it was offering my opinions as a patient on the website, in reviewing the content or adding to the content of the website* *I wanted to make it very meaningful for our patients. So that when they went on this website, it wasn't just information and knowledge, but it was something that they could take away and make it applicable to their everyday life. I felt like I really was able to help achieve that goal.* *I think that because I had more…as a pharmacist, I have more or a great deal of information about drugs that I was able to contribute. Because of my background doing research of clinical trials, I was able to explain to the team and to add and contribute to the development of the website by describing the use of the clinical trials.gov website and identifying potential studies that people might want to volunteer for* *The focus group, I just thought went so well, especially when we started to talk about sexuality. You know, I just saw the people open up so much and so much emotion came out in that. Just seeing some of our patients being able to finally have an outlet to talk to people I thought was just really, really good.* *Research and helping with selecting the instruments used to measure the outcome I contribute more in terms of the quality-of-life measurement because that is more my field. My field is pharmacoeconomic and patient related outcomes, quality of life outcomes*
	• Male perspective	*I think the other way that I contributed was because I'm a male and there aren't a lot of males who have the disease, or a lower percentage of males have the disease. But I was able to provide information about the perspectives from a male perspective.*
	• Spanish speaker	*At that time, it was mostly that we needed some Spanish speaking people in the program. I said, I could understand Spanish, and I could speak the Spanish*
Benefits of involvement	• Engagement with research process • Gains in knowledge of the disease • Acceptance • Help others with the disease • Saw gaps at local level and began filling them • Transference of experience in an academic process to personal business	*The whole process, the meetings that we attended. I learned more about how doctors have to deal with so much and things that a patient really never thinks about. The researchers too. That was interesting to learn what they're doing. Kind of like, I got a behind the scenes view that a regular patient wouldn't see.* *Selfishly, the education. I loved it because you force fed it to me and you really shoved it down my throat, which was great. I needed that., I think the education was tremendous because for the first time ever, I was forced because I was part of this wonderful group, I was forced to look at everything about scleroderma from A to Z. So, there was a lot that I kind of said oh, this doesn't really pertain to me so forget it in the past and in this particular case, I couldn't. And then of course, the other thing that I learned is I learned that no two cases of scleroderma are the same.* *You can't help that you have a disease. So, you have to accept what you have and it's the acceptance I guess, that I could say that they [Project*

(continued)

Table 2. (continued)

Themes	Subthemes	Illustrative quotes
		PIs] helped quite a bit. The acceptance that you got from attending these meetings, and well also to the talks that were given by [PI] and [PI] and stuff like that.
		When some scleroderma patient goes to this website for the first time, and they get reassured that there are solutions and there are options. I feel like, wow. It's kind of cool that I was a small, small, small, small part of that in helping people.
		Well, since I've never had scleroderma before and now, I have scleroderma, the idea to help other people that have scleroderma and not to be so afraid of it like we were. Like 30 years ago when everything that you heard about scleroderma, you knew you would by dying very soon.
		Walked away seeing what a gaping hole we have. Not only as a Michigan chapter but as a larger conversation. And what I was able to do from that is do a training for our support group leaders for that whole topic about what we needed to do more in Michigan to help our patient population. And then from that, I have started to go out to individually reach out to patients and have some individual conversations with some of our patients that I know just getting to know them more on an individual basis and having some one-on-one time with them.
		And again, because I never finished college, some of the things that I saw there, I've actually begun to apply within my businesses. So, with me normally, it's a shoot, ready, aim kind of thing. Let's get it done. Let's get it done today. Let's get it done now. When you come into the world of academics, it's more of…okay, it's created. Let's look at it. Let's tear it apart. Let's rebuild it. Let's create it. Let's relook at it. I think that taught me a lot. You know what? It doesn't matter if I deliver a product to my client in three months or nine months. Just make sure it's thorough. So, I think the thoroughness of the whole process was just an eye-opening experience to me and I've kind of changed my management style a little bit in that sense.
Challenges	• Travel	*I don't travel. The meetings were out of the area and when they thought that we had to go to Michigan, I knew that I couldn't go there. It was during the winter time and definitely, I was not going.*
	• Reviewing the publication manuscripts	*The worst part for me was the reading and the responsibility I felt that A, I had to read it and B, I had to look to perfect it. This one I felt way too much responsibility as a non-academic.*
	• Reviewing the TOSS materials	*Least, I didn't enjoy reading the huge paper. I didn't mind reading like when it was a little section of content, that it was broken up. But when we got that paper…I don't remember which paper it was, but it was about 14 pages. I was like, oh my.*
Project leadership	• Genuine involvement of patient partners in all aspects of the project by key leaders and their staff	*The fact that I'm here and I'm working among people of such professionalism, and everyone treats everyone with such dignity and such respect. I mean, the people in this team are brilliant doctors, scholars and just people that are coming from different walks of life. But yet, everyone is an equal in the work that they're doing and that has been just…I mean, I don't even know that there is a word I can put to it because it has just been amazing to me to see this. That [PIs] mean, these are brilliant doctors that are…and yet, they treat me like a colleague and what an amazing gift that they've given to me to share their expertise. That is truly humbling to be among people of their gifts and talents and allowing me to share that and that has just been such a gift to be in meetings and collaborating with these people. Being allowed to share that has just been very humbling. It's just been such a rewarding experience.*
		Like in the beginning, I kind of felt like oh, I'm just a little patient here. These guys are big, you know, big time. Way smarter than me, that's for sure. But they…oh, everybody made me feel so comfortable and

(continued)

Table 2. (continued)

Themes	Subthemes	Illustrative quotes
		like I could share. Sometimes, [one PI] would personally ask the patients like, what do you think? What do you think? He was very aware that we might be afraid to speak up. So, he really made us feel comfortable to speak up.

from the beginning and then the assembly of all parts of the website (eg, narration, visuals, and resources) was informative. *Acceptance,* another subtheme, was developed from direct involvement in the project as patient partners learned more about SSc and their own disease as well. A patient partner commented "it [TOSS] gives me hope." *Help others with the disease* related to feelings expressed by both patient partners and stakeholders when reviewing the original online version and revised versions of different modules, giving feedback, and being part of the "evolution." One participant said [TOSS] is "without a doubt, the most all-encompassing, reassuring website out there. There are solutions and options …" Another said TOSS was the "one place shopping" to go for credible information. Within the subtheme of *gain knowledge*, patient partners expressed that by co-facilitating focus groups and reviewing the modules in TOSS, they learned more about the disease that benefited them individually, especially techniques to manage their own symptoms. Stakeholders also gained more knowledge of the disease as well as the impact on people's lives. The subtheme, *saw gaps at local level and began filling them,* was voiced by the stakeholders. They recognized the need for the foundations to inform medical professionals, people with SSc and their families about unmet needs of people with SSc, such as intimacy and sexuality. In addition, all participants expressed the need to inform others about the existence of the TOSS website. *Transference of experience in an academic process to personal business* revolved around changes made after participating on the research team. One participant mentioned changing management styles at work. Another, who was also a researcher, commented on the different research style used in the study in regard to patient engagement and the focus on what is of importance to patients with SSc.

The theme, "**Challenges**" referred to challenges shared across several patient partners in regard to travel and by both patient partners and stakeholders in the amount of reading material. We did have 2 face-to-face meetings and participants were invited to be part of a poster presentation at a patient conference. All members of the research team reviewed at least 2 modules for the TOSS website for content and usability. As all participants were included as authors on abstracts and other publications, they were invited to review those prior to submission. Regardless of the demand on their time, the final theme *"**Project Leadership**,"* revealed feelings of belonging and "being truly valued by project leaders" expressed by all participants.

Discussion

Themes that emerged in regard to contributions, benefits, and challenges were similar to findings from other studies that assessed engagement from the perspective of the patient partners and stakeholders (1,6-11).

The lived experience of being a person with SSc was voiced as major contribution to the project. Similar to findings from other studies, as knowledgeable experts, the patient partners identified outcomes that were important to them and others with SSc, and both patients and stakeholders actively participated in the study protocols, especially developing content that was useful and needed for people with SSc. Benefits to participants were also similar to those reported in other studies as participants gained knowledge of the research process, and contributed to helping others (6-10).

However, we also found different benefits such as acceptance, gains in knowledge, and helping others. These were perhaps because SSc is a rare condition and many people, as well as health care providers, have not heard of SSc; information on the internet can be inaccurate or frightening (18). The knowledge and confidence reported by the patient partners agrees with Hamilton's review (10) where participants stated they learned more about their diseases and helping others.

Although not obvious from the comments, the authors, as researchers in the TOSS study, observed that the perspectives of the patients and stakeholders were instrumental in determining outcome measures, and stratifying participants for the randomized control trials (16). Patients and stakeholders developed powerful testimonials, reviewed content, developed resources, participated in the usability analysis of the TOSS website, and co-facilitated focus groups (16). Similar to other studies, we found that time, travel, and cost for the participants to attend research team meetings were challenges (6-10). Even though these costs to patient partners and stakeholders were included in the grant budget, reimbursement was challenging due to institutional policies regarding non-university employees. In addition, because patient partners and stakeholders were part of the research team, co-facilitated focus groups, and were privy to findings from the study, our institution required them to complete human subjects training and most training is written at the college level.

Our research team was geographically dispersed across the United States, so the majority of our meetings were online. We only had 2 face-to-face meetings and a couple of participants presented at the national patient's conference; all costs associated with travel and lodging were covered by the grant. Travel can be a challenge for people with mobility problems, lung and GI issues, and fatigue, which are common with people with scleroderma (19). The recent pandemic created a new common venue of videoconferencing and method to conduct meetings which is especially useful to engage patient and stakeholder partners as research team members. Other challenges, not mentioned in previous studies, were onerous reading and reviewing of the website and publications. However, they enjoyed reviewing the website. Perhaps, future studies could offer patients and stakeholders summarized versions in lay terms of final reports and publications, in addition to the more formal versions of final reports and publications; feedback could be requested on the lay term version.

Participants reported feeling supported and valued as members of the team and that their opinions mattered which is in contrast with findings from other studies. The rareness of SSc has created a tight community as patients, family members, patient organizations, researchers, and clinicians are invested in improving quality of life for those with SSc.

Limitations

Interviews were conducted at the end of the project and relied on memories over a 3 plus year timespan. Future studies might want to interview patients/stakeholders at different points in time or after different phases during a research study. However, as stated earlier, our goal was to obtain perspectives over the lifetime of the project as it seemed people gained more perspectives and were engaged in different stages of the research as the study time went on. Future studies might want to obtain input from the rest of the research team in regard to their perspectives of benefits and challenges of patient and stakeholder engagement. Another limitation, that could have created bias and affected the responses to the interview questions, was that the interviewer and PIs were known to the participants. Using an interviewer not familiar with the research team might have yielded different results.

Conclusion

Patients and stakeholders engaged in the same research study reported on their contributions, and benefits and challenges similar to those expressed in other studies. Additional benefits were identified: acceptance, increased knowledge of SSc, and helping others. Moreover, positive experiences with the research team leadership were expressed. The findings support the contribution of patient partners and stakeholders as key members of the research team in identifying key outcomes, and helping set the tone of interviews. Importantly, they provided insights based on their experiences that improved and enhanced the relevance of the final product, TOSS.

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Supplemental Material

Supplemental material for this article is available online.

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