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DOI
10.46439/Psychiatry.1.014

Publication date
2021

Document Version
Final published version

Published in
Current Research in Psychiatry

Current Research in Psychiatry

Heijerman-Holtgrefe et al., Curr Res Psychiatry 2021; 1(3):40-43.

Commentary

Active involvement of patient representatives in research: roles, tasks, and benefits in a pilot intervention study

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Received date: May 04, 2021
Accepted date: July 19, 2021

Abstract

Patient involvement in research and development has been increasingly applied and studied during the last decades. There is rising consensus that collaboration with patients or their representatives (e.g., parents, family members, partners) in co-developing new treatment interventions and participating in treatment studies is beneficial for patients, researchers, and society at large. In studying the feasibility of *Tackle your Tics*, an innovative treatment program for children with tic disorders, patient representatives played a unique role throughout the research process, performing multiple important tasks from start (co-designing the study, obtaining a grant, developing and leading workshops and parent meetings) to finish (interpretation of the data, providing feedback to the article, giving congress oral presentations). This commentary paper offers a brief research note providing tangible examples of the roles, tasks and benefits of patient representatives in research. Active involvement of patient representatives in the *Tackle your Tics* pilot study has provided improvements in the treatment program with experience-based knowledge, and improvements in the research design, matching the needs and wishes of participating families. These benefits of involving patient representatives can be relevant for future research designs.

Keywords: Patient involvement, Patient perspective, Experience-based knowledge, Tourette syndrome, Tic disorders

Introduction

Traditionally, patients' involvement in research has been limited to providing data as research subjects, recruiting research participants, or funding research. Most of the research design and activities were conducted by academically educated researchers only. In recent decades, a shift has been made towards a greater influence of patients with the aim to better meet their needs and priorities [1-3]. With patients on a higher step on the 'participation ladder' [4], their role can vary from advisor to full research partner, initiator, and equal member within the research team. Furthermore, parents, relatives, and partners of patients can be involved in research, based on their experiential knowledge of how to cope with tics and other problems in daily life with tic disorders. In this paper, patient representatives are defined as patients or their parents, relatives, and partners, who share their experience-based knowledge to improve research and the quality of care from the patient's perspective.

In a recent literature review on the 'return on patient engagement initiatives', Vat et al. summarized various benefits of patient participation for research and development in usability, obtaining funding, design, and ethics. For example, by incorporating experience-based knowledge, research can be more relevant and usable for patients and be better attuned to patients' needs. It can contribute to the quality of research because of a more appropriate, inclusive and sensitive research design, improved recruitment, adherence, and satisfaction by participants [5]. Crocker et al. indicated that patient and public involvement (PPI) improves enrolment of participants, especially if it includes people with lived experience of the studied condition [6]. Recently, a Dutch study into 'social return on investment' (SROI) demonstrated that peer support is of great benefit to patients and society, leading to higher quality of life, less absenteeism, higher productivity, and a shorter care process, thereby reducing costs [7].

Equal partnership in mixed research teams includes involvement in all research activities from beginning to end, with a focus on experiential knowledge, openness, respect, and mutual learning, which helps to overcome stereotypes [8]. The Dutch national patient organization (Stichting Gilles de la Tourette, Tourette.nl) is a non-profit, volunteer driven organization. It has approximately 1,000 members and 50 active volunteers. Its aim is to inform people about Tourette Syndrome, other chronic tic disorders and comorbidities, to provide peer support, and engage in advocacy on behalf of those living with the condition and their families.

For over ten years, the organization has an established and equal collaboration with clinicians and researchers in the organizations' scientific advisory board. This long collaboration between experts with academical, clinical, and experience-based knowledge was the base for studying the feasibility of a new treatment program for children and adolescents with tic disorders, called *Tackle your Tics* [9].

This commentary aims to outline the roles and tasks of patient representatives in this feasibility study. The benefits of active involvement of patient representatives for developing and studying a new intervention program can be relevant for future research designs.

Roles, Tasks, and Benefits

Tackle your Tics was developed by a team consisting of experienced clinicians (child and adolescent psychiatrists and psychologists), researchers, and patient representatives (adult patients and parents). Two patient representatives are active volunteers and (former) board members of the patient organization. Author AH was appointed to the research team as PhD-student, to conduct the pilot study (in preparation of a large multicenter randomized controlled trial). Author LB was responsible for introducing, monitoring, and safeguarding the patient perspective in the research process. Additionally, patient representatives were involved as study participants, focus groups participants, workshop leaders, and mentors in parent meetings. See Table 1 for an overview of their roles and tasks, which will be described in detail.

Participants

In September 2018 and February 2019, 14 children and adolescents with tic disorders were included in a feasibility study into a brief intensive behavioral therapy program, *Tackle your Tics*. In this program, participating children (9-17 years) were offered behavioral therapy (Exposure and Response prevention for tics) according to the 'Tics' protocol by Verdellen et al. [10] in small groups during four days. Motivating elements were added to the program: workshops on coping strategies given by young adult patients and trained patient representatives, a training app, parent meetings, psychoeducation, and relaxation exercises. Participants and parents were asked to complete online interviews and questionnaires pre- and post-treatment and at 2 months follow-up, to study treatment outcomes and treatment satisfaction.

The children and adolescents participated in two groups of respectively 6 and 8 children and adolescents with tic disorders at Levvel, Academic Centre for Child and Adolescent Psychiatry and Specialized Youth Care in Amsterdam. *Tackle your Tics* appeared to be a feasible therapy program for reducing the severity of the tics and improving quality of life, with high treatment satisfaction [9]. On the treatment satisfaction questionnaires (parent and child version), the mean score was almost 4 at the five-point scale questions (Table 2). Due to the findings of this small pilot, a multicenter randomized controlled trial with more than 100 participating children and adolescents is in process.

Advisors

During the study preparation phase, a patient advisory board of parents and young adults with a tic disorder was installed, to provide feedback on the study and program design. Based on their input, some adaptations were applied to the program and research process: (1) the researchers clarified information and psycho-education to guard against unrealistic expectations of the treatment outcomes (e.g., a total disappearance of tics) and misunderstandings; (2) questions were added to the treatment satisfaction questionnaires, e.g., to check

Table 1: Overview of roles and tasks of patient representatives in *Tackle your Tics*. Based on: Abma et al. [8].

Role	Task
Participant	Participating in the treatment program as patient or parent; completing interviews and questionnaires
Advisor	Participating in the focus group on the research design before the start of the pilot study; joining the follow up feedback meeting for parents
Counselor	Providing program elements: workshops, parent meetings, and psychoeducation
Research partner/initiator	Initiating research, jointly developing the design, grant applications, recruitment, data gathering and analysis, publications, and congress presentations, joining research networks

Table 2: Treatment satisfaction scores (N=13) of program components provided by the patient representatives on a 1-5 scale questionnaire, child version (11-items) and parent version (22-items) (1=not helpful at all, 5=very helpful).

	Children			Parents		
Treatment satisfaction scores	**mean**	**range**	**SD**	**mean**	**range**	**SD**
total score	3.94	3.27-4.91	0.456	3.92	2.48-4.64	0.559
psycho-education	4	1-5	1.080	4.23	2-5	1.092
workshops	4.08	3-5	0.641	3.82	2-5	0.874
parent meetings				3.92	2-5	0.760

the need for more care or support after the program; and (3) all assessments were completed online in the home environment instead of during the treatment program, to build a positive and safe group atmosphere during treatment.

After the treatment and all measurements were completed, a follow-up meeting was organized. Participating children and parents were reunited to exchange experiences and provide feedback. Parents attended a presentation of the pilot results by the researchers, at which they were asked for feedback. During this presentation, the children had a relaxing time in an indoor playground. One parent reported their child felt no necessity or obligation to practice the home exercises anymore after the last treatment day, which was confirmed by other parents. Another parent felt she had few tools to help her child with the exercises. The input of this meeting and the findings from the treatment satisfaction questionnaire were used to further improve the program in follow up research: (1) adding parent sessions, in which parents can learn to help their child during exercises at home and (2) an extra follow-up meeting after one month to encourage children to practice.

Counselors

Two adult patients and trained patient representatives, both licensed BSc-level educational professionals, provided psychoeducation in collaboration with an experienced cognitive behavioral therapist. In addition, they developed and provided workshops on coping with tics and other symptoms. In these experience-based workshops, participating children learned from young adult patients and role models how to cope with their symptoms in a positive mindset, focusing on positive characteristics and strengths.

An experienced support group counselor and patient representative provided the parent meetings, together with one of the therapists. Parents were able to exchange experiences and support each other in coping with problems at home, in school or in society. On the treatment satisfaction questionnaires designed for this study (parent and child version), the program components provided by the patient representatives were all assessed positively (Table 2). In open questions, several children and parents reported that they felt supported.

Research Partners/Initiators

The pilot study was initiated by the scientific advisory board of the Dutch national patient organization Stichting Gilles de la Tourette. Patient representatives were already part of the initiating research team from start to finish and therefore actively involved in all research phases, based on shared decision making. They participated in the development of the design, grant applications, recruitment, data gathering and analysis, publications and congress presentations, and joining research networks.

In the study design, as in the majority of studies in this field, tic reduction was seen as the key outcome for determining the efficacy of the intervention. Patient representatives emphasized a broader view, stressing the importance of quality of life as an equally (if not more) important outcome. Therefore in our design and statistical analyses (repeated measures ANOVAs from pre- to post-assessment to follow-up), we decided to use and consider quality of life as our main secondary outcome next to the key outcome tic severity, from pre- to post-assessment to follow-up.

Patient representatives also emphasized the need for improved treatments that are more accessible and with less burden for children and families. In accordance with a large European patient survey [11], the patient organization stated that treatment should not focus on tics only but also on other symptoms, characteristics, and personal strengths, as well as on coping with symptoms in daily life and with additional peer support.

The pilot study was funded by a partner patient organization, Tourettes Action in the UK, with the Research Grant Award in 2018. The patient involvement aspects of this study were presented by patient representatives at international congresses [12].

Discussion and Conclusions

The *Tackle your Tics* study is a positive example of equal partnership of academic researchers, clinicians, and patient representatives, in a mixed research team, across all research phases and activities. The teams' aim was to continue and optimize this collaboration in a follow up randomized controlled trial. Where patient involvement is often set up for strategic reasons (e.g., to obtain funding or recruitment), this was not the case in this study. There is a shared vision in the team that patient involvement improves research in a way that is indispensable for the main goal of this research: improving the treatment and daily lives of children with tic disorders and their families.

Involvement of trained patient representatives in this pilot study delivered several important benefits in the treatment development and research process. First of all, it added valuable experience-based knowledge to the treatment program, which helped children and families to cope with tics and related problems in their daily lives. In addition, involvement of patient representatives ensured that the treatment program and the research design and process better matched the needs and wishes of participating families.

Although the examples and benefits described in this paper can be relevant for future research designs, we identify some limitations for generalizing these findings. First of all, our team had a long history of cooperation in the patient organizations' advisory board and therefore most team members were familiar with working together with patient representatives. The positive results and benefits may not be representative for studies with newly formed teams, in which the roles and tasks of patient representatives have yet to be defined, which may cause challenges. Vat et al. [5] summarize challenges for different research partners and organizations, found in other studies. For example, patient representatives can experience confusion due to a lack of clarity about roles and procedures or disappointment due to a mismatch of expectations. Researchers can face methodological concerns and costs, as well as stress due to new ways of working together with patients and advocacy groups, which can be associated with competence or power struggles. Second of all, in this small pilot study, we did not perform a standardized evaluation of the involvement of patient representatives, for example as described in the article of Boivin et al. [3].

Based on our findings, we can conclude active involvement of patient representatives in the *Tackle your Tics* pilot study has shown important benefits, which can be relevant for future research designs. Consideration of possible challenges, and the monitoring and evaluation of the cooperation process are recommended for other mixed research teams.

Acknowledgements

The authors thank Tourettes Action in the UK for their Research Grant Award in 2018 (Grant number 1003317), which funded this study, the participating children and families, clinicians, and volunteers of the patient organization (Dutch Tourette Association) for their valuable contributions, the Dutch Tourette Association for funding additional publication costs, and Marlon Schotel, MA, for additional proofreading.

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