


Patient and public involvement in oesophageal cancer survivorship research

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ABSTRACT

Background: Good clinical research is often conducted in close collaboration between patients, the public, and researchers. Few studies have reported the development of patient and public involvement (PPI) in research outside the United States and the United Kingdom, and for patients with more aggressive cancers. The study aimed to describe and evaluate the development of PPI in oesophageal cancer survivorship research in Sweden by the use of a framework to support the process.

Methods: Oesophageal cancer survivors were recruited to a PPI research collaboration at Karolinska Institutet, Sweden. The development process was supported by the use of a framework for PPI, 'Patient and service user engagement in research'. Insights, benefits, and challenges of the process were described and discussed among the collaborators.

Results: The collaboration resulted in joint publications with a more patient- and family-focussed perspective. It also contributed to the development of information folders about survivorship after oesophageal cancer surgery and national conference arrangements for patients, their families, health-care workers, and researchers. Since the PPI contributors were represented in patient organisations and care programmes, the dissemination of research results increased. Their contributions were highly valued by the researchers, but also revealed some challenges. The use of a structured framework contributed to support and facilitated the process of establishing PPI in research collaboration.

Conclusions: A genuine interest in establishing PPI in research and an understanding and respect for the patients' expertise in providing a unique inside perspective was imperative for a successful collaboration. Research focus should not only be on mortality and reductions in daily life, but also on positive outcomes. Using a framework supports development and avoids pitfalls of PPI collaboration.

Patient and public contribution: Patient partners were equal collaborators in all aspects of the study.

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Patient and public involvement; oesophageal neoplasm; cancer survivorship; theoretical model

Background

Patient and public involvement (PPI) in research is defined as research activity carried out 'with or by' members of the public rather than 'to', 'about', or 'for' them [1]. In PPI, the public is an active partner in one or several parts of the research, for example, in generating research questions, study design, analysis, evaluation/interpreting of the results, co-writing manuscripts, and dissemination of the publications [2]. Involving patients and the public's perspectives in health care research suggests an increase in the quality and value of the research, more efficient implementation of results, and subsequently health care improvements [3–8]. The National Health Service in the United Kingdom (UK) states that research becomes more relevant to patients, is robustly conducted, and better communicated if using PPI and strongly encourage researchers to involve patients and the public in their research [9]. In a recent publication, it was stated that the effectiveness of PPI was strongest when people experienced in living with the disease were involved as research

partners, based on the view that patients and the public have complementary knowledge to that of scientists [10]. Most studies on PPI originate from the United States, the United Kingdom, and Australia, and focussed on patients with less aggressive and advanced cancer diseases [11]. In this study, we established a PPI collaboration within oesophageal cancer survivorship research in Sweden. Oesophageal cancer is the seventh most commonly diagnosed cancer worldwide [12] and carries a poor prognosis with an overall 5-year survival below 20% [13,14]. The majority of patients are elderly (>60 years) men [15]. Curative treatment commonly involves extensive surgery (oesophagectomy), chemotherapy, and sometimes radiotherapy [15] which may, in turn, negatively impact survivorship. Many survivors experience life-long problems with an altered life situation [16] and reduced health-related quality of life [17]. Since this debilitating disease influences patients' lives long after the end of curative treatment, the survivorship research should focus on what is valuable for the individual patient and how health care can help patients live meaningful lives.

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No study has reported the use of structured PPI in oesophageal cancer survivorship research. Therefore, this study aims to describe and evaluate the development of PPI in oesophageal cancer survivorship research in Sweden by the use of a framework to support the process.

Framework for PPI

In this study, we used a framework to facilitate the development of PPI collaboration. Our choice was based on a systematic review of 41 theoretical frameworks and conceptual models for PPI in research and synthesised into one new pragmatic framework for standardising the structure of patient-research collaboration, entitled: 'Patient and service user engagement in research' [18]. The first part of the framework involved the establishment of the PPI and consisted of four parts:

1. *Patient and service user (PPI) initiation.* There is a strong emphasis on early inclusion in the process, which will enable the PPI contributors to influence the content of the research projects and provide input on study design, applicability, and ensure a relevant perspective. The PPI contributors should consist of individuals for whom the outcomes are of interest. There must be a potential for them to take an active role in the collaboration and agreement of expectations.
2. *Building reciprocal relationships.* PPI contributors and researchers must value each other as equal partners of the research team. Well-defined roles and responsibilities facilitate this process. The collaboration should include a mutual understanding of the partners' needs, capacities, and goals.
3. *Co-learning.* Education and training about content or methodology are imperative to carry out a productive dialogue. Co-learning may increase PPI contributors' confidence, promoting more active engagement and reducing the risk of tokenistic participation. Also, researchers must be taught about introducing participatory collaboration.
4. *Re-assessment and feedback.* The PPI contributions and expectations should be continuously evaluated and modified when needed. Identified mistakes can be corrected and used in improving the framework.

When the collaboration is established, the collaborators are faced with three phases of research work; the preparatory-, the execution-, and the translational phase. In the preparatory phase, agenda-setting and funding are essential activities. The execution phase includes the conductance of the study and completing the data collection. The translational phase is the post-analysis phase and consists of activities such as dissemination and implementation of results and evaluation of the work cycle.

Methods

Design

This was a descriptive study presenting the development process of a PPI collaboration within oesophageal cancer survivorship research supported by a structured framework.

Context

The Surgical Care Science research group at Karolinska Institutet, Stockholm Sweden, conducts research on cancer survivorship after oesophageal cancer surgery, mainly by using patient-reported outcome measures. In 2016, a collaboration of mutual interest between researchers and patients was initiated. The main purpose was to inform and guide the research through patients' experiences, but also for the research to provide support for the patients through a better understanding of the disease. The establishment of the collaboration was supported by the 'Patient and service user engagement in research' framework [18]. A steering group consisting of the principal investigator, a research nurse with close patient contact, and a researcher experienced in patient engagement work was established. The group decided how to recruit participants for the PPI collaboration, based on the PARADIGM recommendations [19].

Participants

PPI contributors were primarily recruited through an ongoing prospective longitudinal study on survivorship after oesophageal cancer surgery entitled: Oesophageal Surgery on Cancer patients – Adaptation and Recovery (OSCAR) [20]. Participants were advertised for using social media, the research centre's web page, and the webpage of a patient organisation for pancreas, liver, gastric, and oesophageal cancer (palema.org). Factors that were considered important for inclusion were a genuine research interest and social and collaborative skills. The individuals should represent the patient population diversity regarding age, gender and ethnicity. All applicants received a comprehensive information folder about what to expect from the collaboration. They were later contacted by email or telephone to verify their interest and were invited to join the collaboration. The final group was composed of an equal number of researchers and PPI contributors. The PPI contributors were mostly elderly men, but also some women and younger people. A certain PPI contributor turnover was anticipated, but 6 to 10 PPI contributors in the collaboration was the goal.

Procedure

The initial meetings were dedicated to identifying and discussing the purpose of and setting the format for the collaboration. The PPI contributors had an active role in establishing the platform, but apart from that, they did not receive any formal education or other preparations to facilitate the collaborative work. All meetings were held at the research office, outside of the hospital, to avoid the potential emotional stress that the hospital environment could evoke. In-person meetings were considered to be preferable in order to build a trustful relationship between the team members. All meetings included a coffee break with a snack since malnutrition is a common problem after oesophagectomy because of the post-surgical changes in anatomy [21]. The official meeting language was Swedish – the mother tongue

of the PPI contributors. However, to be able to understand scientific talks by English-speaking researchers, all PPI contributors had to understand English. Economic compensation for attendance was proposed but declined by the PPI contributors except for reimbursements for travel costs. No specific funding was allocated for the collaboration and the costs were included in the daily expenses. The meetings were held four times/year, lasted approximately 2 hours, and had flexible agendas in case important discussions arose. On the agenda typically, were presentations of research ideas, data collection, and grant applications. However, common for all meetings were discussions regarding the ongoing prospective longitudinal study OSCAR [20]. To start with, the researchers presented ongoing and planned research projects to the PPI contributors for their input. They provided their perspectives on the studies and on how to improve the quality of the research to ensure that the outcomes were important to oesophageal cancer survivors. The PPI contributors also helped with the interpretation of the results (from their perspective). Some of them were researchers in fields other than medicine and they suggested original research ideas. Together, researchers and PPI contributors designed and conducted common studies, some of which have been published.

Evaluation

The applicability of the framework in establishing a PPI collaboration was discussed in the group and yearly meetings about how to improve the collaboration were held. Meeting notes and other initial documents were reviewed. Subjective opinions, anecdotes, and lessons learned were reported in the results. The guidance for reporting of PPI in health and social science research (GRIPP) checklist was used where applicable to assure the quality, transparency, and consistency in the manuscript writing [22]. The researchers and the PPI contributors have co-authored the present manuscript together.

Results

Table 1 summarizes aspects of the collaboration, described in relation to the 4 initial parts of the framework.

Table 1. The PPI contributors' and the researchers' perspectives on important aspects of the collaboration, described in relation to the 4 initial parts of the framework.

The 4 parts of the framework	PPI perspective	Researcher perspective
PPI initiation	Sometimes doubted the representativeness of own opinions	Selection of individuals for the PPI collaboration was crucial
Building reciprocal relationships	Lack of medical knowledge, vocabulary, and research Fear of ending up in a position of inferiority among the researchers	Creating a confident atmosphere in the group Emphasizing that all participants have equally important roles in the work process
Co-learning process	With time they felt increasingly confident and motivated to learn more	Each meeting included a lecture about a certain research field that could be understood by laymen
Re-assessment and feedback	The discussions sometimes induced emotionality	The collaboration required special efforts, time, and patience

PPI: patient and public involvement.

Patient and service user (PPI) initiation

Researchers' comments:

Careful identification of individuals for the PPI collaboration was essential. The patient choice cannot only be selected based on the registration of interest by patients. Researchers must assess the person's physical and emotional conditions and the ability for participation as well. Being a reflective individual with a genuine interest in research was probably more important than an academic education. PPI contributors with previous experience from research facilitated the start-up of the collaboration.

PPI contributors' comments

Motivations to be involved in medical research may vary. Some PPI contributors were driven by altruistic motives – to contribute to research that would ultimately improve survivorship and quality of life after surgery. Others were driven by a desire to talk about their cancer, care, and recovery to persons who were interested and prepared to listen. Some had a general interest in the care after surgery and the possibility to learn things that would benefit them in their own recovery or had a general curiosity and interest in the research process. However, it was difficult to know if the PPI contributors' opinions were representative of other patients and if their experienced problems were related to the disease or to the fact that they were ageing or retired.

Building reciprocal relationships

Researchers' comments:

Based on the knowledge that trust and mutual respect are important factors for a successful collaboration [23], great efforts were put into ensuring a confident atmosphere in the group that would make the PPI contributors feel comfortable. Coffee breaks were important for the group to get to know each other on an informal basis. Further, special attention was given to emphasise that all participants had complementary and equally important roles in the work process. During the scientific discussions, it was continuously confirmed that the PPI contributors understood all important details. A research nurse coordinating the meetings helped in bridging the gap of knowledge and potential jargon barriers between the group members. All PPI contributors'

suggestions and advice were taken seriously and helped set out directions for research. In cases when the researchers chose different pathways, they always motivated their choice.

PPI contributors' comments

Lack of medical knowledge and vocabulary, and a general lack of knowledge about research were factors that were perceived as barriers for becoming part of a research group. Some PPI contributors feared that they would end up in a position of inferiority among the professional experts in the group. Sometimes, they were hesitant to speak on behalf of other patients in the same situation, with doubt on how representative their own experiences and symptoms were. The researchers encouraged all inputs in the discussions, independent of perceived subjectivity. It was stated that all members represented themselves and it was up to the researchers to use the information selectively.

Co-learning process

Researchers' comments:

Altruistic reasons may not be enough to motivate the PPI contributors to stay in the research group in the long run. Therefore, each meeting included a lecture or presentation about a certain research field (e.g., psychological effects of cancer, nutritional problems, and common statistical methods) followed by a discussion to improve the individual meaningfulness. Here, the PPI contributors could engage in the discussions and contribute with their expertise to the scientific discussions. All new PhD students presented their research projects in the group to discuss the significance of the project from a PPI contributor perspective. Exchange students highly appreciated these meetings since they improved their understanding of the survivors' situation and the relevance of their projects.

PPI contributors' comments

The presentations during the meetings were on relevant topics and were well prepared on a level that could be understood by laymen but still not trivial. The researchers were eager to answer any questions in a friendly but still professional way. With time the PPI contributors felt increasingly confident and motivated to learn more. The seminars were also a good opportunity for the PPI contributors to exchange experiences about their recoveries.

Re-assessment and feedback

Researchers' comments

Feedback on the collaboration was regularly provided during meeting discussions and was used to improve the work. The degree of involvement in the research, from the generation of new ideas to evaluation of results and review of manuscripts, required special efforts and patience from the researchers as well as from the PPI contributors. Potential

new group members were also discussed to achieve good representativeness for oesophageal cancer patients.

PPI contributors' comments

The PPI contributors experienced that it was possible to discuss research in detail with the researchers. However, the collaborative work was also burdensome since the discussions sometimes induced emotionality. Talking in detail about their cancer experience was sometimes uncomfortable and they found themselves wandering between recognition (subjectivity) and objectivity, for example, while feedbacking on draft articles. Sometimes the knowledge generated anxiety and brought up personal problems that were not previously part of the daily worries. A certain degree of anxiety had to be accommodated and they emphasised that discussions should not always be focussed on symptom burden, deterioration, and mortality, but also on more positive findings such as recovery and improving quality of life.

The preparatory, execution and translational phases

Researchers' comments

In the first papers, the PPI contribution was mentioned in the acknowledgements [24–29]. After a while, they started to identify their own research ideas and became more involved in drafting manuscripts, and were also included as co-authors, like in the present paper. So far, they have had input on most of the published studies based on data from the OSCAR cohort. For example, the PPI contributors suggested the outline for the first paper describing the study cohort with a paper focussing on common problems experienced by patients in the first year after surgery [20]. Currently, 10 papers from this cohort have been published and a report including a summary of findings from these studies have been released and distributed among the study participants. Since many of the remaining problems after oesophageal cancer surgery are related to food intake, the PPI contributors suggested a study on attitudes towards eating and eating habits, which led to the initiation of a research collaboration with a restaurant school. Another study originated from a discussion about prognosis where the PPI contributors recognised the need for more information on prognosis to have a realistic plan for future life. This resulted in a prediction model on postoperative survival after oesophageal cancer surgery using register-based data which was published in a high-impact journal [29]. This study would not have been conducted without input from the PPI contributors. During one meeting, the idea of developing a leaflet about the long-term effects of oesophageal cancer surgery came up, since there was a lack of such information. Relevant data were collected, assessed, prioritised, and compiled to an information folder directed to patients with oesophageal cancer and their families. The content and language of the folder was confirmed to be relevant and understandable by the PPI contributors and was published online. Another initiative was the arrangement of an oesophageal cancer survivorship conference for patients, family members,

researchers, and health care workers. The PPI contributors participated in the planning phase, development of the programme, and one individual was invited as a speaker at the conference. The event rendered positive feedback from those who attended. For example, 95% stated that the conference contributed to an increase in their knowledge about recovery after surgery for oesophageal cancer. On the PPI contributors' initiative, the conference was followed up with a folder containing information on the presentations.

PPI contributors' comments

Some of the PPI contributors were involved in a patient organisation of oesophageal cancer, which enabled the dissemination of recent research findings in a way that the researchers were unable to do. The patient organisation presented new research findings on their webpage, on seminars or webinars, reaching out to the intended target group of the research. The PPI contributors were also found as representatives in care programmes and cooperation with clinicians from the hospital, excellently bridging the gap between patients, clinicians, and researchers.

Discussion

The collaboration resulted in patient- and family-focussed publications co-authored by the PPI contributors (so far, one study has been published) [30], the development of post-surgery information folders, and conference arrangements. Also, dissemination of results improved since the PPI contributors were represented in patient organisations and care programmes and could help in spreading news about how to improve quality of life long after the end of treatment.

Previous studies have reported benefits of patient engagement in research, such as in a research priority setting [31,32], in the design of clinical studies [33,34], and the regulatory processes [35,36]. The major challenges of these studies seem to include time and costs for planning and managing the collaboration [6]. However, during the establishment of PPI collaboration in oesophageal cancer survivorship research, we identified some other important factors for a successful collaboration:

1. A genuine interest in establishing a collaboration with the patients together with a sincere understanding and respect that the patients are experts on providing a unique inside perspective is crucial.
2. Biases in the recruitment of patients and risks of ending up with a homogenous sample not representative for the studied group have previously been highlighted [37]. This was noted as a problem in this study as well, where the current group consisted of a restricted number of highly educated individuals who probably were not well representative of the whole population of surgically treated oesophageal cancer patients. However, experiences of functions and symptoms following oesophageal cancer surgery, which was the focus of our

research, are probably not dependent on education, social status, ethnicity, or sex.

3. To reduce the risk of awakening feelings of distress among the PPI contributors, meetings should not only be focussed on mortality and reductions in daily life, but also on positive outcomes.
4. Attempting to develop PPI for long-term research collaborations instead of an individual research project, increased the complexity, but the PPI contributors may find a long-lasting relationship more valuable to invest their time and efforts in. However, if there is no new recruitment, the participants may be distanced from the event with time and not updated on the latest treatments.
5. Using a framework helps to guide the development of the PPI collaboration. For us, it made the process more effective and avoided some preventable obstacles. The first part of the framework was particularly helpful in identifying how our context enabled or restrained patient engagement and made us aware of the wide range of potential the collaboration could imply. The second part was less feasible since it was more directed towards PPI in a specific research project and not as a whole.

Some limitations that need to be considered in this report are that the framework was originally intended to support reporting of PPI activities and not specifically for the development process of PPI collaboration such as in the present study. Still, the framework was pragmatic and flexible and proved to be useful in establishing a PPI collaboration. Moreover, the evaluation was based on subjective experiences by few individuals and meeting notes. No assessment tool for measuring efficacy or user satisfaction was used. Therefore, some valuable reflections might have been missed. However, since we were able to establish a well-functioning PPI collaboration, it suggests that the framework helped with directions and advice on what was important in establishing a well-functioning PPI collaboration.

Most likely, the experiences and lessons learned from using this framework in establishing a PPI collaboration with patients who survived such a severe disease as oesophageal cancer are probably attainable in other less ill patient groups and transferrable to similar research settings.

Conclusions

PPI in research improves the patient relevance of the research, as well as facilitating the dissemination of research findings. Lessons learned are that a sincere interest in establishing PPI in research, as well as an understanding and respect for the patients as experts on providing a unique inside perspective, is imperative for the collaboration. The focus on research should not only be on mortality and reductions in daily life, but also on positive outcomes. Furthermore, the use of a supportive framework may provide rigour and structure to the development process and be helpful in avoiding pitfalls.

Disclosure statement

The authors declare that they have no competing interests.

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Data availability statement

Data sharing does not apply to this article as no datasets were generated or analysed during the current study.

References

- [1] Turk ABA, Locock LA. Researcher's Guide to Patient and Public Involvement. Oxford 2017 [cited 2021 6 September]. Available from: <https://oxfordbrc.nihr.ac.uk/wp-content/uploads/2017/03/A-Researchers-Guide-to-PPI.pdf>.
- [2] Price A, Albarqouni L, Kirkpatrick J, et al. Patient and public involvement in the design of clinical trials: an overview of systematic reviews. *J Eval Clin Pract*. 2018;24(1):240–253.
- [3] INVOLVE. Briefing notes for researchers: Involving the public in NHS, public health and social care research 2012. [cited 2021 Jan 28].
- [4] Hoven E, Eriksson L, Mansson D'Souza A, et al. What makes it work? Exploring experiences of patient research partners and researchers involved in a long-term co-creative research collaboration. *Res Involv Engagem*. 2020;6:33.
- [5] Domecq JP, Prutsky G, Elraiyah T, et al. Patient engagement in research: a systematic review. *BMC Health Serv Res*. 2014;14:89
- [6] Brett J, Staniszewska S, Mockford C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. *Health Expect*. 2014;17(5):637–650.
- [7] Vat LE, Finlay T, Jan Schuitmaker-Warnaar T, et al. Evaluating the "return on patient engagement initiatives" in medicines research and development: a literature review. *Health Expect*. 2020;23(1): 5–18.
- [8] Gradinger F, Britten N, Wyatt K, et al. Values associated with public involvement in health and social care research: a narrative review. *Health Expect*. 2015;18(5):661–675.
- [9] National Health Research Authority. Public involvement: The National Health Service England; 2021. Available from: <https://www.hra.nhs.uk/planning-and-improving-research/best-practice/public-involvement/>.
- [10] Crocker JC, Ricci-Caballo I, Parker A, et al. Impact of patient and public involvement on enrolment and retention in clinical trials: systematic review and Meta-analysis. *BMJ*. 2018;363:k4738
- [11] Pii KH, Schou LH, Pii K, et al. Current trends in patient and public involvement in cancer research: a systematic review. *Health Expect*. 2019;22(1):3–20.
- [12] Sung H, Ferlay J, Siegel RL, et al. Global cancer statistics 2020: GLOBOCAN estimates of incidence and mortality worldwide for 36 cancers in 185 countries. *CA A Cancer J Clin*. 2021;71(3): 209–249.
- [13] Kauppila JH, Mattsson F, Brusselaers N, et al. Prognosis of oesophageal adenocarcinoma and squamous cell carcinoma following surgery and no surgery in a nationwide swedish cohort study. *Bmj Open*. 2018;8(5):e021495.
- [14] Office for National Statistics. Cancer survival in England - adults diagnosed 2019. [Jun 27, 2021]. Available from: <https://www.ons.gov.uk/peoplepopulationandcommunity/healthandsocialcare/conditionsanddiseases/datasets/cancersurvivalratescancersurvivalinenglandadultsdiagnosed>.
- [15] Lagergren J, Smyth E, Cunningham D, et al. Oesophageal cancer. *Lancet*. 2017;390(10110):2383–2396.
- [16] Scarpa M, Valente S, Alfieri R, et al. Systematic review of health-related quality of life after esophagectomy for esophageal cancer. *World J Gastroenterol*. 2011;17(42):4660–4674.
- [17] Schandl A, Lagergren J, Johar A, et al. Health-related quality of life 10 years after oesophageal cancer surgery. *Eur J Cancer*. 2016;69:43–50.
- [18] Shippee ND, Domecq Garces JP, Prutsky Lopez GJ, et al. Patient and service user engagement in research: a systematic review and synthesized framework. *Health Expect*. 2015;18(5):1151–1166.
- [19] Innovative Medicine Initiative. Patient engagement in medicines development: Recommendations on how to find the right match for the right patient engagement activity: Patients active in research and dialogue for an improved generation of medicines (PARADIGM); 2020. [cited 2021 April 23]. Available from: <https://imi-paradigm.eu/PEtoolbox/identification-of-patient-representatives.pdf>.
- [20] Schandl A, Johar A, Anandavadevelan P, et al. Patient-reported outcomes 1 year after oesophageal cancer surgery. *Acta Oncol*. 2020;59(6):613–619.
- [21] Martin L, Lagergren J, Lindblad M, et al. Malnutrition after oesophageal cancer surgery in Sweden. *Br J Surg*. 2007;94(12): 1496–1500.
- [22] Staniszewska S, Brett J, Simeria I, et al. GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research. *Res Involv Engagem*. 2017;3:13.
- [23] Ludwig C, Graham ID, Gifford W, et al. Partnering with frail or seriously ill patients in research: a systematic review. *Res Involv Engagem*. 2020;6:52.
- [24] Lagergren P, Johar A, Liu Y, et al. Severe reflux and symptoms of anxiety and depression after esophageal cancer surgery. *Cancer Nurs*. 2021. DOI:10.1097/NCC.0000000000001026
- [25] Liu Y, Pettersson E, Schandl A, et al. Psychological distress after esophageal cancer surgery and the predictive effect of dispositional optimism: a nationwide population-based longitudinal study. *Support Care Cancer*. 2021;5(1):1–7.
- [26] Schandl A, Malberg K, Johar A, et al. Social distancing during the COVID-19 pandemic and health-related quality of life among esophageal cancer survivors. *Acta Oncol*. 2021;60(8):1011–1018.
- [27] Liu Y, Pettersson E, Schandl A, et al. Higher dispositional optimism predicts better Health-Related quality of life after esophageal cancer surgery: a nationwide Population-Based longitudinal study. *Ann Surg Oncol*. 2021;28(12):7196–7205.
- [28] Liu YJ, Schandl A, Markar S, et al. Psychological distress and health-related quality of life up to 2 years after oesophageal cancer surgery: nationwide population-based study. *BJS Open*. 2021; 5(1):zraa038.
- [29] Xie SH, Santoni G, Malberg K, et al. Prediction model of long-term survival after esophageal cancer surgery. *Ann Surg*. 2021; 273(5):933–939.
- [30] Lagergren P, Johar A, Rosenlund H, et al. Severe reflux, sleep disturbances, and health-related quality of life after esophageal cancer surgery. *J Cancer Surviv*. 2021;15(6):818–824.
- [31] Geissler J, Ryll B, di Priolo SL, et al. Improving patient involvement in medicines research and development: a practical roadmap. *Ther Innov Regul Sci*. 2017;51(5):612–619.
- [32] Duffett L. Patient engagement: What partnering with patient in research is all about. *Thromb Res*. 2017;150:113–120.
- [33] Ray KN, Miller E. Strengthening stakeholder-engaged research and research on stakeholder engagement. *J Comp Eff Res*. 2017; 6(4):375–389.

- [34] Esmail L, Moore E, Rein A. Evaluating patient and stakeholder engagement in research: moving from theory to practice. *J Comp Eff Res.* 2015;4(2):133–145.
- [35] Borup G, Bach KF, Schmiegelow M, et al. A paradigm shift towards patient involvement in medicines development and regulatory science: Workshop proceedings and commentary. *Ther Innov Regul Sci.* 2016;50(3):304–311.
- [36] Levitan B, Getz K, Eisenstein EL, et al. Assessing the financial value of patient engagement: a quantitative approach from CTTI's patient groups and clinical trials project. *Ther Innov Regul Sci.* 2018;52(2):220–229.
- [37] Blackburn S, McLachlan S, Jowett S, et al. The extent, quality and impact of patient and public involvement in primary care research: a mixed methods study. *Res Involv Engagem.* 2018;4:16.