From Department of Learning, Informatics, Management and Ethics
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# Supportive resources for self–care and informal care – Uncovering the role of patient–driven innovations

Marie Dahlberg

Karolinska Institutet

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# Supportive resources for self-care and informal care – Uncovering the role of patient-driven innovations
# Thesis for Doctoral Degree (Ph.D.)

By

# Marie Dahlberg

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**Principal Supervisor:**
Associate Professor Carolina Wannheden
Karolinska Institutet
Department of Learning, Informatics, Management and Ethics
Medical Management Centre

**Co-supervisor(s):**
Ami Bylund, PhD
Sophiahemmet University & Karolinska Institutet
Department of Learning, Informatics, Management and Ethics
Medical Management Centre

Professor Petter Gustavsson
Karolinska Institutet
Department of Clinical Neuroscience
Division of Psychology

Teresa Herlestam, PhD, Oncologist & Palliative Care Specialist
ASIH Aleris & Regional Cancer Centre Stockholm-Gotland

**Opponent:**
Professor Sofia Kjellström
Jönköping University
Department for Quality Improvement and Leadership
School of Health and Welfare

**Examination Board:**
Professor Anette Alvariza
Marie Cederschiöld University
Department of Health Care Sciences
Palliative Research Centre

Professor Susanne Guidetti
Karolinska Institutet
Department of Neurobiology, Care Sciences and Society
Division of Occupational Therapy

Professor Axel Wolf
University of Gothenburg
Institute of Health and Care Sciences
Centre for Person-Centered Care (GPCC)

To Simon and Vera, my constant and loving reminders of what life is all about

In memory of Rickard

# Popular science summary of the thesis

In recent decades, technology has advanced rapidly, leading to many new health solutions that support individuals in their self-care. More research is being done on such innovations, which are often developed and driven by persons living with illness themselves or by informal caregivers (e.g., family members or friends who generally provide unpaid support). However, we still know little about these patient-driven innovations — like what needs they address, how people use them, and what impact they have on individuals in general and for self-care and informal care in particular. Therefore, the main aim of this thesis was to explore what kinds of support matter to persons with long-term health conditions and their informal caregivers, and how patient-driven innovations can help them in their self-care and in the provision of informal care.

The thesis consists of four studies. The study participants in the first two studies were persons living with brain tumors and informal caregivers in Sweden. Living with brain tumor may cause significant impairment to individuals' physical and mental health, as well as social functioning. Thus, persons living with brain tumors are often dependent on help from informal caregivers. Study participants were introduced to a social network mapping tool called CareMaps, which was developed by an informal caregiver to help individuals to identify relations that matter in their self-care. The first study found that the study participants valued different types of relationships that either supported them in their self-care or helped them to maintain their personal identities disconnected from the illness experience. The second study focused specifically on the experiences of informal caregivers and described how they used different types of resources to balance caregiving with other aspects of life. The third study broadened the perspective by exploring patient-driven innovations that have been published in the research literature. These innovations fulfilled various roles by supporting self-care, open sharing of information and knowledge, and enhancing collaboration with healthcare. However, few studies have evaluated their impact for the persons using them and for healthcare. The fourth study explored why patient innovators choose to publish their work and their experiences with the publication process. Many published to strengthen the patient voice in research and had both positive and negative experiences to share. The findings highlight the need for ongoing support for patient innovators and other patients and members of the public to contribute their experiences and knowledge to create meaningful research and services that support self-care and caregiving.

# Populärvetenskaplig sammanfattning

Under de senaste årtiondena har tekniken utvecklats snabbt, vilket har lett till många nya hälsoinnovationer som stödjer individer i sin egenvård. Mer forskning bedrivs om sådana innovationer, som ofta utvecklas och drivs av personer som själva lever med sjukdom eller av anhöriga (till exempel familjemedlemmar eller vänner som generellt sett ger stöd utan ersättning). Dock vet vi fortfarande lite om dessa patientdrivna innovationer — som vilka behov de adresserar, hur människor använder dem, och vilken påverkan de har på individer generellt och för egenvård och anhörigvård specifikt. Därför var huvudsyftet med denna avhandling att utforska vilka typer av stöd som är viktiga för personer med långvariga hälsotillstånd och deras anhöriga, och hur patientdrivna innovationer kan hjälpa dem i sin egenvård och i tillhandahållandet av anhörigvård.

Avhandlingen består av fyra studier. Studiedeltagarna i de första två studierna var personer som lever med hjärntumörer och anhöriga i Sverige. Att leva med en hjärntumör kan orsaka betydande funktionsnedsättningar som påverkar individers fysiska och mentala hälsa, samt sociala funktion. Därför är personer som lever med hjärntumörer ofta beroende av hjälp från anhöriga. Studiedeltagarna introducerades till ett verktyg för kartläggning av sociala nätverk (CareMaps), som utvecklats av en anhörig för att hjälpa individer att identifiera relationer som är viktiga för deras egenvård. Den första studien fann att studiedeltagarna värderade olika typer av relationer som antingen stödde dem i deras egenvård eller hjälpte dem att bibehålla sin personliga identitet frikopplad från sjukdomssituationen. Den andra studien fokuserade specifikt på anhörigas upplevelser och beskrev hur de använde olika typer av resurser för att balansera anhörigvård med andra aspekter av livet. Den tredje studien vidgade perspektivet genom att utforska patientdrivna innovationer som har publicerats i forskningslitteraturen. Dessa innovationer fyllde olika funktioner genom att stödja egenvård, möjliggöra fri delning av information och kunskap, och förbättra samarbete med vården. Dock har få studier utvärderat deras effekter för personerna som använder dem och för vården. Den fjärde studien utforskade patientinnovatörers upplevelser av publiceringsprocessen. Många publicerade för att stärka patientens röst inom forskningen och hade både positiva och negativa erfarenheter att dela med sig av. Resultaten understryker behovet av fortsatt stöd för patientinnovatörer och andra patienter och allmänheten för att bidra med sina erfarenheter och kunskaper för att skapa meningsfull forskning och tjänster som stödjer egenvård och anhörigvård.

# Abstract

**Background**: Persons living with long-term conditions and informal caregivers are often dependent on support for their self-care and informal care. The experience of insufficient support may lead some to develop health innovations to address their unmet health needs (i.e., patient-driven innovations). Although research on patient-driven innovations is increasing, knowledge about the needs that such innovations address, how and by whom they are used, and their outcomes is still limited. Empirical studies are needed to understand the potential benefits and challenges of patient-driven innovations for self-care, informal care, as well as health service delivery. Further, the role of patient innovators in health services research merits investigation.

**Aim**: The overall aim of this thesis was to explore which supportive resources matter to persons living with long-term conditions and informal caregivers and how patient-driven innovations can help facilitate self-care and informal care. The aim was addressed by exploring patient-driven innovations in different contexts. Study I explored the use of a caregiver-developed social network-mapping tool (CareMaps) to assess quality of social and healthcare relations. Study II explored how such relations could be used as supportive resources for self-care and informal care. Study III explored the objectives and outcomes of patient-driven innovations that have been published in peer-reviewed journals. Study IV explored patient innovators' reasons for and experiences of authoring scientific publications about their innovations.

**Methods**: Four qualitative studies were conducted. Studies I and II were conducted in the context of brain tumor self-care and informal care in Sweden. Study I was an interview study with persons living with brain tumors, informal caregivers, and bereaved caregivers, and collected data were analyzed using thematic analysis. Study II was an interview study with informal caregivers of persons living with brain tumors, and collected data were analyzed using a combination of conventional and directed content analysis. Study III was a content analysis of scientific publications that were included in a previously published scoping review of patient-driven innovations. Study IV was an interview study with international patient innovators from three continents who had published in scientific journals. Collected data were analyzed using the Framework Method.

**Findings**: Study I found that persons living with brain tumors, informal caregivers, and bereaved caregivers expressed positive opinions about using the CareMaps tool but raised some questions regarding its design and how best to use it in their self-care and informal care. Self-care supportive relations and identity-preserving relations emerged as two distinct types of relations that participants valued. They were found in different contexts and emphasized contrasting qualities. Study II found that informal caregivers combined various resources both to manage emotional distress related to caregiving and to make space for valued activities and relationships disconnected from caregiving. In Study III, 83 publications covering 21 patient-driven innovations were analyzed. The patient-driven innovations illustrated a diversity of innovative approaches to facilitate daily lives of persons living with long-term conditions and informal caregivers, interactions with peers, and collaborations with healthcare. Few publications reported on outcomes. Most of the innovations have been developed for use on an individual or community level without healthcare involvement. Study IV found that patient innovators engaged in scientific publishing primarily to strengthen the patient voice in research and to gain recognition for their innovations. Although they had positive experiences of research and publication processes, they also faced cultural and structural barriers, such as conservative peer review practices and publications behind paywalls.

**Conclusions**: This thesis indicates that persons living with long-term conditions and informal caregivers are central stakeholders in driving health service development and research forward to meet the needs that matter to persons living with long-term conditions and informal caregivers. The findings elucidate that continued efforts are needed to facilitate for patient innovators, as well as other patient and public contributors, to contribute with their experiences and expertise to the production of relevant and meaningful research and services supporting self-care and informal care.

# List of scientific papers

I. **Dahlberg M**, Bylund A, Gustavsson P, Herlestam Calero T, Wannheden C. What matters to persons living with brain tumors and their informal caregivers? An interview study of qualities in interpersonal relations. Social Science and Medicine, 2022;292:114575-114575

II. **Dahlberg M**, Wannheden C, Andersson S, Bylund A. "Try to keep things going" – Use of various resources to balance between caregiving and other aspects of life: an interview study with informal caregivers of persons living with brain tumors. Manuscript.

III. **Dahlberg M**, Lek M, Malmqvist Castillo M, Bylund A, Hasson H, Riggare S, Reinius M, Wannheden C. Objectives and outcomes of patient-driven innovations published in peer-reviewed journals: a qualitative analysis of publications included in a scoping review. BMJ Open, 2023;13:e071363

IV. **Dahlberg M**, Luckhaus J.L, Hasson H, Jansson H, Lek M, Savage C, Riggare S, Wannheden C. Why publish? An interview study exploring patient innovators' reasons for and experiences of scientific publishing. Submitted to Research Involvement and Engagement, 26 January, 2024

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# Prologue

I grew up in a large family. Although I am an only child, my childhood was filled with people: my parents, my maternal grandparents, my aunt and teenage uncles, and our German Shepherd, Rinty, my first best friend. It was a clearly defined, loving, supportive and solid unit – my first experience of a social group.

My interest in the impact of social relations on people's health and well-being began during my degree in Global Studies at the University of Gothenburg's Faculty of Social Sciences. For my master's thesis (D-level) on the involvement of men in the fight against HIV/AIDS, I conducted a minor field study in Kampala, Uganda. Here, it became evident to me how crucial social support is for sharing similar experiences, for providing comfort, preventing isolation, and fostering awareness and competence to mitigate risk behavior. Following these studies, I dedicated ten years to working on similar issues at a Swedish NGO, Noaks Ark. Here, I witnessed first-hand how peer support positively influenced individuals' well-being, serving as a source of inspiration and motivation for self-care (even though I was not acquainted with the term at the time).

Throughout my life, the number of social groups and families to which I belong has grown. In each of these groups I have found support during times of triumph and adversity, joy, and sorrow. When Rickard, my beloved husband, passed away in 2012 after a brief battle with an aggressive cancer, I was left to care for our children Simon and Vera, who were eight and six years old at the time. Our lives were turned upside down. Looking back, we recognize how all our social groups, in different ways and to different extents, mobilized to support us, unconditionally, guiding us toward a new normal. For that, we are profoundly grateful!

When I first encountered CareMaps, it sparked my interest on both a personal and a professional level. This social network mapping tool was designed to help individuals identify supportive relations. It was not the first of its kind, but it was unique in that it was developed by a person with lived experiences as an informal caregiver – a patient innovator. This perspective on healthcare and self-care intrigued me. When the opportunity arose to delve into the phenomenon of patient-driven innovations – exploring their utility and the motivations that led individuals to share their innovations for the benefit of others – I seized it.

# 1 Introduction

More and more people are living longer lives. Improved living conditions, earlier diagnosis, and detection of illness, advancing medical treatments and preventive interventions, and innovative digital health technologies have increased longevity (1-3). At the same time, ageing populations, unhealthy lifestyles, and environmental risk factors (e.g., air pollution) have increased the number of people living with long-term conditions (4, 5). In 2019, 36% of the adult population in the EU, and 38% in Sweden, were living with at least one long-term condition (6). These individuals often require long-term care management and holistic, tailored support for self-care (7, 8). Meanwhile, many countries are facing a workforce crisis in healthcare coupled with budget cuts to public healthcare spending (9, 10). The rising demand for healthcare services due to demographic shifts and the rising prevalence of chronic illness, coupled with workforce shortages, inadequate working conditions, and strained resources, place immense strain on healthcare professionals and on healthcare systems (11-13). Taken together, these developments underscore the importance of effective self-care strategies. Collaborative efforts, policy reforms, and innovative self-care and informal care support are vital to address these challenges and ensure better health outcomes for individuals living with long-term conditions (9, 14).

Meanwhile, people living with long-term conditions and informal caregivers engage in daily routines and activities to balance the impact of the illness situation with other aspects of life, such as being a parent, partner, or co-worker. They may make regular healthcare visits for check-ups or treatment follow-ups, but spend most of their time engaging in self-care or informal care without healthcare involvement (15). In response to insufficient or inaccessible support tools and services, persons living with long-term conditions and informal caregivers may develop their own solutions (i.e., patient-driven innovations) to facilitate daily life. The Patient Innovation website (www.patient-innovation.com), which gathers and shares patient-driven innovations, listed over one thousand innovations in February 2024. This thesis explores patient-driven innovations in the context of brain tumor self-care and informal care in Sweden, and in the wider context of research publications in peer-reviewed journals.

The thesis was conducted within the research programs "Co-care for persons living with long-term illness" (2018-2020) and "Patients in the driver's seat!" (2021-2024), both funded by the Swedish Research Council for Health and

Welfare (Forte), and the Swedish Research Council. The aim of the Co-care program was to develop, implement, and evaluate models of partnership care (i.e., co-care). The concept of co-care stresses that the role of healthcare providers is to complement individuals' own resources to achieve the best possible health outcomes (16). Rather than involving patients in healthcare, healthcare should be involved in patients' and informal caregivers' self-care, based on their individual goals and preferences. The Patients in the driver's seat is a partnership program that builds on patient-driven innovations to promote self-care and co-care and studies their implementation into everyday health services and the daily lives of patients. This thesis builds on one of the innovations that was included in both research programs, the CareMaps tool, which is a pen-and-paper tool for visualizing social relations and healthcare resources and aims to support individuals in their self-care, informal care, and collaboration with healthcare. Figure 1 provides an overview of the studies.
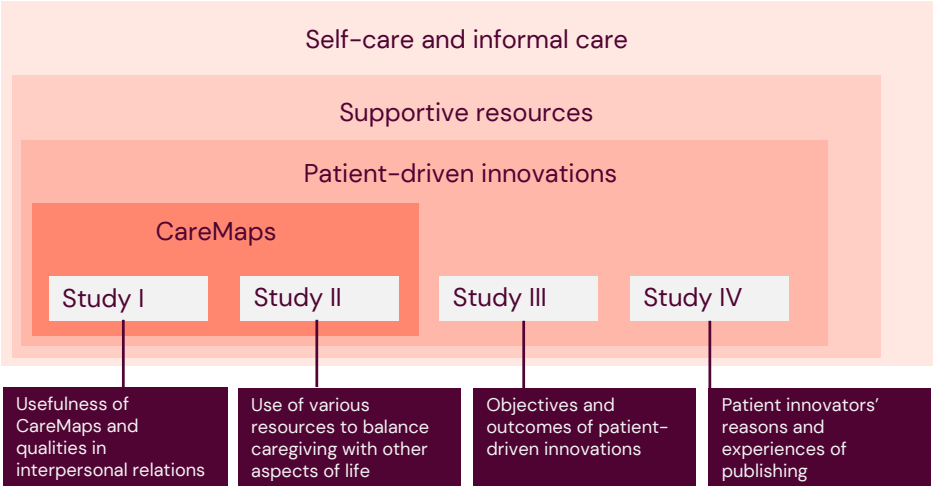


Figure 1. Overview of the studies.

# 2  Literature review

## 2.1  Self-care

Beyond hospital visits for treatment and follow-up, individuals with long-term conditions spend most of their time outside healthcare settings, actively engaging in self-care**.** There are several definitions of self-care (17). In this thesis, I will use the WHO definition, which states that "self-care is the ability of individuals, families and communities to promote health, prevent disease, maintain health, and to cope with illness and disability with or without the support of a health-care provider" (18). In healthcare, self-care refers to the ability to manage disease symptoms, treatments, and related consequences, including lifestyle adjustments (19). Thus, self-care involves a dynamic interplay between individual abilities, external support, and proactive management, to ensure overall health and well-being.

Self-care practices include three key components: *self-care maintenance*, which involves maintaining emotional and physical stability, whether ill or healthy; *self-care monitoring*, whereby individuals routinely observe changes in symptoms and signs related to their health, which helps them stay informed about their condition; and *self-care management*, which entails actively managing physical and emotional symptoms as they occur, and can include following specific autonomous or healthcare-recommended interventions, seeking appropriate care, or adjusting lifestyle choices and routines (20, 21). Additionally, people living with long-term conditions are increasingly contributing to improvements in healthcare, drawing upon their self-care knowledge and personal experiences to enhance healthcare processes and services. Their insights play a valuable role in shaping better healthcare outcomes (22, 23).

Practicing self-care depends on an individual's capability to make informed choices, exercise judgment, and take proactive steps to address their own needs, and assess individual and environmental resources to maintain well-being – referred to as self-care agency (20, 24). Despite what the term may imply, self-care is also a team effort. In addition to self-care agency, the ability to engage in self-care relies on the provision of healthcare support and social support.

## 2.2 Supportive resources to self-care

### 2.2.1 Healthcare support for self-care

Healthcare professionals play an important role in providing holistic support to ensure effective self-care (20). Self-care support programs, timely information and communication, and individually tailored support (e.g., counselling and training) are essential to empower patients, enhance their understanding of diagnoses, and improve coping abilities (25–30). Recent decades have seen an increase in digital health tools (e.g., smartphone applications and wearables) that enable communication, increased participation, and shared decision-making in care and treatment processes and in self-care (1, 31). Recognizing the importance of self-care and tailoring strategies to individual needs can lead to better health outcomes and improved well-being (32, 33). Thus, self-care interventions offer promising health benefits, but also involve new working methods, which require training and capacity building of healthcare staff to equip them with the necessary skills and ensure provision of appropriate self-care support (8). To enhance provision of self-care support to individuals living with long-term conditions, several frameworks to offer guidance for system-wide changes have been developed (14). Recommended strategies for self-care support include patient education programs, training and awareness building among healthcare professionals, and digital health technologies for self-care management.

### 2.2.2 Informal caregiving

Outside healthcare, self-care often involves family members, relatives, friends and other social support resources, who play an important role in providing informal self-care support (20). An informal caregiver is defined as an individual who "provides – usually unpaid – care to someone with chronic illness, disability or other long-lasting health or care need, outside a professional or formal framework" (34). Informal caregiving includes various levels of instrumental support; for example, assisting with household duties, medication, and care coordination, as well as providing emotional and social support (35). In Sweden, approximately 20% of the adult population, of which 80% are of working age (18–64 years), provide informal care and support for an average of thirteen hours per week (36). In cases where less time is spent on actual caregiving tasks, informal caregivers may still spend many hours on "stand-by" (37).

Caregiving can be a rewarding experience, leading to a sense of social connectedness and enhanced personal growth and self-esteem, and reduced stress and depressive symptoms (38–40). More commonly, however, informal caregiving is associated with caregiver burden, which has been described as "the level of multifaceted strain perceived by the caregiver from caring for a family member and/or loved one over time" (41). Caregiver burden can include experiences of physical and psychological burden including stress, anxiety, or depression (42–45). The higher the caregiver burden, the greater the risk of decreased health and quality of life, especially among caregiving spouses and partners (46). There is also research encouraging a more balanced view of caregiving, where positive and negative aspects and experiences are not mutually exclusive, but multidimensional, forming a continuum of experiences from stress to satisfaction (38, 47, 48).

Informal caregiving is often undertaken with little or no support or training provided, and balancing caregiving with household, family, work, and social activities can be a struggle (42, 49, 50). In addition, the transition from an intellectually and functionally equal relationship to a context predominated by illness and caregiving can be challenging (51, 52). The informal caregiver may experience a shift from being a partner or a child to the person concerned, to gradually becoming consumed by caregiving at the expense of other self-identities (e.g., that of being a parent or colleague) and shared identities (e.g., that of a partner) to a level where caregiving becomes the "master identity" (43).

As informal caregivers to persons living with long-term conditions are a critical part of the care ecosystem, providing essential support to those in need, it has been argued that they are not just a complement to health- and social care but constitute a separate level of care (34, 53).

### 2.2.3 Social relations, social networks, and social support

Social relations are defined as recurring and interpersonal connections perceived to have personal meaning to those involved (54). Having social relationships is vital for individuals' health and well-being, and may reduce mortality (55). Conversely, a lack of social relationships is a risk factor comparable to smoking, alcohol abuse, physical inactivity, and poor diet (55). Social networks, a defined set of social relationships, are a social determinant of health and may have a positive or a negative influence on an individual's choices and behavior and thus, on health (54, 56, 57). Social networks can be

characterized by structure (size and density), interaction (frequency and reciprocity), and function (types of support, development of new social relations, and preservation of social identity) (58). While quantity of social relations is measured by number and frequency, the main quality dimension of social relations and networks is social support (59).

A social network is a prerequisite for, but does not guarantee, social support (60). Certain characteristics are necessary for social support to occur: social embeddedness, the depth and strength of the relations within the social network providing an environment of protection and helpfulness (i.e., social climate); and reciprocity, the mutual exchange of resources between at least two people (56, 60, 61). Reciprocity has been shown to have a positive impact on health; it moderates stress (62), depressive symptoms (63), and enables caregiving and self-management (64). Social capital is a term used to describe the accumulated benefits of potential and actual resources that are gained from participating and investing in a social network (65). Social capital can be objective or subjective (62). Objective social capital is enacted support that is actually provided when needed. Subjective or perceived social capital is the sense of belonging to a group or community and the belief that support is available if needed, thus promoting a sense of well-being (62). Subjective support is predictive of well-being, while objective support is critical when actually needed (66). For example, informal caregivers who perceive having available support may feel less burdened regardless of the actual amount of support provided (67).

### 2.2.4   Coping resources

Living with a long-term condition or being an informal caregiver to a person living with long-term condition can be stressful, and the ability to cope with stress is necessary to maintaining health and well-being. Stress can be defined as change, and whether it is valued as positive or negative depends on the individual's reaction to the event or challenge causing stress (i.e., the stressor) (68). According to Folkman and Lazarus' transactional model of stress and coping, coping can be understood as a process that involves both cognitive and behavioral responses to manage stressors (i.e., external or internal demands) that are perceived to exceed the person's resources (69). Coping arises from the appraisal of stressors, involving primary appraisal (i.e., assessing whether the stressful situation implies harm, threat, or challenge) and secondary appraisal

(i.e., assessing whether the situation can be controlled). When resources are insufficient to control the situation, stress can arise either directly from the objective burden of the stressful event (e.g., illness progression; cognitive, behavioral, or physical condition) or from the subjective perception or burden (e.g., assumption of responsibilities and obligations) (70). Coping with stress involves management of the main stressors (problem-focused coping) and regulation of emotional distress (emotion-focused coping) (71).

Carver, Sheier and Weintraub developed the multidimensional COPE inventory to assess the different ways in which individuals respond to stress beyond problem-focused and emotion-focused coping (72). The authors argued that the difference between the responses to stress may have various implications for an individual's coping success. The COPE inventory was later revised and condensed into the Brief COPE consisting of 14 dimensions (active coping, planning, positive reframing, acceptance, humor, religion, using emotional support, using instrumental support, self-distraction, denial, venting, substance use, behavioral disengagement, self-blame) (73). The Brief COPE has been widely used to assess responses to challenging life events (74-79).

Resources required for coping can be categorized into intrapersonal resources (e.g., own experiences, personality characteristics, strengths, and coping skills) (80), interpersonal resources (family, friends, or community), and professional resources (healthcare or other professional services) (75, 81). Interpersonal (social) support is critical for coping. A stable partner, family, and friends increase the use of adaptive coping strategies, and in turn, adaptive coping strategies enhance the perception of social support (79, 82). Having several social support resources beyond informal caregivers, including wider social networks and new friendly relationships, encourages active and adaptive coping (79).

### 2.2.5 Social network mapping

There are several tools available to assess social resources. Different types of social network mapping can be used to identify social relations and networks for social support. Social network mapping has been used in anthropology to study how individuals interact with each other in different social contexts (83). In epidemiology, it is used as a tool to identify and understand the spread of infectious diseases (84). In healthcare settings, social network diagrams are used to visualize social relationships and assess supportive resources beyond next of

kin (85, 86). As social networks are unique to each individual, mapping them provides a context in which to identify and navigate existing and potential resources for patients, including informal as well as formal caregivers (preventive care, healthcare, or social care) (87, 88).

One example of social network mapping is ecomapping, which helps to understand individuals in the context of a complex ecological system going beyond individual health concerns to consider broader social contexts (89). It was originally developed in the 1970s for the assessment of family needs in public child welfare practice (89). Ecomapping has previously been used for data collection, for example in nursing practice to assess care support needs (90, 91) and in nursing research on family caregiving among individuals and groups (92–94). The use of ecomapping may have a facilitating effect on the relationship between the interviewer and the interviewee, which may lead to disclosure of information that otherwise may not have been shared in an interview situation (91, 95).

Atlas CareMaps™ is an ecomapping tool that was developed specifically for informal caregivers of persons living with long-term conditions (88, 96). It has been described as "a tool and process for understanding family care ecosystems" by "sparking transformative conversations" and has been shown to contribute to informal caregivers' self-reflection, understanding of their own degree of dependence on caregiving and support outside formal care, and improved communication with healthcare professionals (96, 97).

## 2.3   Co-production

Recent decades have seen a shift in philosophy of care towards empowering and actively involving persons living with illness and informal caregivers in service delivery through collaborative designing and decision-making (98). Patient-centered care and person-centered care are different but interrelated concepts currently dominating the healthcare agenda. Both involve a patient-professional relationship defined by empathy, respect, engagement, communication, individualized holistic focus, shared decision-making, and collaborative care coordination over time (98). The major difference is that patient-centered care is applied in clinical decisions, whereas person-centered care focuses on the person beyond their medical conditions, taking into account the significance and impact of mental, emotional, and social needs (99).

Whether implemented individually or collectively, both concepts serve to strengthen the role of persons living with illness as co-producers of health. Co-production is a service delivery approach designed to impact on service users through involvement of the users themselves and the development of collaborative partnerships in healthcare (100). It has been described as an exploratory and reflective process of engagement and interaction that challenges the traditional patient–professional relationship in two ways: firstly, it requires skills training for healthcare professionals to help them move from "fixers to facilitators" and adapt to collaboration with service users as experts; and secondly, it demands the competence and skills of service users, including decision-making and responsibility towards the community that they represent (100, 101). Thus, co-production is about the empowerment of both service users and healthcare staff. Individuals are no longer merely care recipients, but co-producers of care; examples include self-management of treatment, self-monitoring for registering and reporting of health data, co-producing individual health and treatment plans, or collaborating on co-production of tools or services (102). Co-production is also used in multidisciplinary collaborations in research to improve research quality, relevance, and its impact on policy and practice, with different stakeholders working together as partners in shaping research agendas, designing and implementing studies, interpreting findings, and disseminating results (103).

### 2.3.1 Patient and public involvement in research

Patient and public involvement (PPI) is a related concept to co-production that is often used in healthcare services research (104). The purpose of PPI is to make healthcare and research more relevant and useful for those directly affected by its outcomes. PPI in research is carried out "with" or "by" – rather than "about" or "for" – members of the public, thus actively involving those it aims to benefit, leading to more impactful and relevant outcomes for everyone involved (104, 105). PPI goes beyond tokenistic participation and fosters meaningful relationships and collaborations between researchers, clinicians, and those impacted by the research (persons living with illness, informal caregivers, and the wider public). By involving patients and the public in influencing research design, data collection, and delivery of findings, research becomes more person-centered, which ultimately leads to better health and social care services, improved treatments, and higher-quality care (106–108). The principle of patient and public involvement is increasingly being embraced and prioritized by the

research community, health stakeholders, scientific journals, regulators, and funders (109-111). To facilitate involvement there are an increasing number of guidelines being published for planning, conducting, and reporting PPI in various contexts and types of research and healthcare partnerships (112-114).

## 2.4  Patient-driven innovations

While the PPI concept suggests that service users are involved in research or development processes initiated by healthcare or public sector organizations, such processes can also be initiated by service users themselves. Patient-driven innovations are where service users become independent innovation sources, driving and developing "innovations either individually or collaboratively within their social systems" to achieve sustainable solutions and services (115). In this thesis, I use the definition of patient-driven innovations as described by Reinius et al. (116). They define "patient-driven" as initiated, developed, and driven by patients and/or informal caregivers, and as responding to one or several unmet needs of the innovator. For the "innovation" part of the concept, they use the WHO definition of health innovation: "an innovation that identifies new or improved health policies, systems, products and technologies, or services and delivery methods that improve people's health and well-being" (117).

The development of solutions by patients for problem-solving and to facilitate everyday life is a common phenomenon, but far more innovations are being developed than disseminated (118, 119). Many patient innovators may not reflect on the value of their innovations to others, and therefore may not consider sharing it or may make active decisions not to diffuse (120). Those who choose to share their innovations are rarely driven by profit. Patient innovators devote their own time and financial resources to developing solutions for their own use and to be shared with others. Rather than protecting their innovation they often share it openly for anyone to use, free of charge (121, 122).

Patient-driven innovations can range from simple solutions to high-tech devices that aim to enhance the user's ability to perform daily routines and activities and to increase their level of independence (116, 123). Studies have shown that patient-driven innovations often either provide affordable and refined alternatives to existing commercial innovations or fill a gap where solutions are lacking to address unmet needs (123, 124). Although research on patient-driven innovations is increasing (116), knowledge about the needs that such innovations address, how and by whom they are used, and their outcomes is still limited.

Empirical studies are needed to understand the potential benefits and challenges of patient-driven innovations for self-care, informal care, as well as health service delivery. Further, the role of patient innovators in health services research merits investigation.

# 3 Research aims

The overall aim of this thesis was to explore which supportive resources matter to persons living with long-term conditions and informal caregivers and how patient-driven innovations can help facilitate self-care and informal care. The aim was addressed by exploring patient-driven innovations in different contexts. Initially, I focused on a patient-driven innovation that was introduced in the context of brain tumor self-care and informal care in Sweden. Subsequently, I broadened my focus to exploring various patient-driven innovations that have been published in scientific literature. Thus, I had the opportunity to develop both specific and broad knowledge about patient-driven innovations. The four studies of the thesis had the following specific objectives:

Study I: To explore how persons living with brain tumors and informal caregivers perceive the potential usefulness of a social network-mapping tool in their self-care; second, to describe the qualities of the interpersonal relations that they map.

Study II: To explore how informal caregivers of persons living with brain tumors use their intrapersonal, interpersonal, and healthcare and community resources to manage challenges related to caregiving and balance caregiving with other aspects of life.

Study III: To gain a deeper understanding of the objectives and outcomes of patient-driven innovations that have been published in peer-reviewed journals, focusing on the unmet needs that patient-driven innovations address, and the outcomes for patients and healthcare that have been reported.

Study IV: To explore patient innovators' reasons for and experiences of authoring scientific publications about their innovations.

# 4  Materials and methods

This thesis consists of four exploratory studies investigating the various roles of patient-driven innovations in relation to self-care and informal care. All studies have a qualitative research design, which is appropriate for collecting and analyzing data that focus on perceptions and experiences of the participants (125). Contextually, the thesis can be structured into two parts. In Studies I and II, a patient-driven innovation was used as a tool to explore which supportive resources matter to persons living with long-term conditions and informal caregivers. In Studies III and IV, publications identified in a scoping review of patient-driven innovations provided a broader context for delving deeper into how such innovations can act as supportive resources for self-care and informal care. Table 1 presents an overview of the method used in all studies.

Table 1. Information about the studies.

|  | Study I | Study II | Study III | Study IV |
|---|---|---|---|---|
| **Focus** | Perceived useful-ness of the Care-Maps tool and qualities of inter-personal relations | Use of various resources to balance caregiving with other aspects of life | Objectives and outcomes of patient-driven innovations | Patient innovators' reasons for and experiences of publishing |
| **Participants/ Data sources** | Persons living with brain tumor (N=7) Informal caregivers (N=6) Bereaved caregivers (N=6) | Informal caregivers of persons living with brain tumor (N=16) | Peer-reviewed publications on patient-driven innovations (N=96) | Patient innovators (N=15) |
| **Data collection** | Non-participant observation Semi-structured interviews | Semi-structured interviews | Articles included in a scoping review | Semi-structured interviews |
| **Time of data collection** | January-December 2019 | August-October 2020 | May-June 2021 | June-August 2022 |
| **Data analysis** | Thematic analysis | Conventional and directed content analysis | Manifest content analysis | Framework Method |

## 4.1 Exploring supportive resources that matter in self-care and informal care

### 4.1.1 Study context

Studies I and II were conducted in the context of brain tumor self-care in Sweden. In 2022, approximately 1450 people (27 new cases per 100,000 people) in Sweden were diagnosed with tumors in the central nervous system (brain and/or spinal cord), accounting for 2% of all cancer diagnoses (126). Brain tumors may cause multiple impairments including physical (e.g., fatigue, pain, imbalance), cognitive (e.g., memory loss, concentration, comprehension), emotional (e.g., anxiety, depression, apathy), perception (e.g., vision), muscle weakness, and communication (127, 128). There are numerous brain tumor types and classifications. The most common type is glioma, which accounts for 70% of the malignant brain tumors (129). The participants in Studies I and II were persons who reported living with malignant glioma (at the time of the interviews) or informal caregivers to persons who reported living with malignant glioma.

### 4.1.2 CareMaps

I had the opportunity to use the CareMaps tool to explore supportive social and healthcare resources that matter to persons living with brain tumors and informal caregivers in their self-care and informal care. The CareMaps tool was initially developed by an informal caregiver to a person living with a brain tumor. The tool shares similarities with other social network-mapping instruments and its design was specifically inspired by the Atlas of Caregiving project which resulted in a social network-mapping tool for informal caregivers of persons living with long-term conditions (88). CareMaps is a pen-and-paper tool that allows the user to visualize their social and healthcare relations (130). Its purpose is to enable persons living with long-term conditions and informal caregivers to identify, value, strengthen and coordinate relationships and resources that they experience as important in their self-care or informal care. An illustration of the initial version of the CareMaps tool is published in Study I. It consisted of: a set of instructions guiding the user through a process of identifying and valuing relationships that matter to them; pens in different colors; and a paper canvas on which a visual representation of social relations is drawn manually (Figure 2).
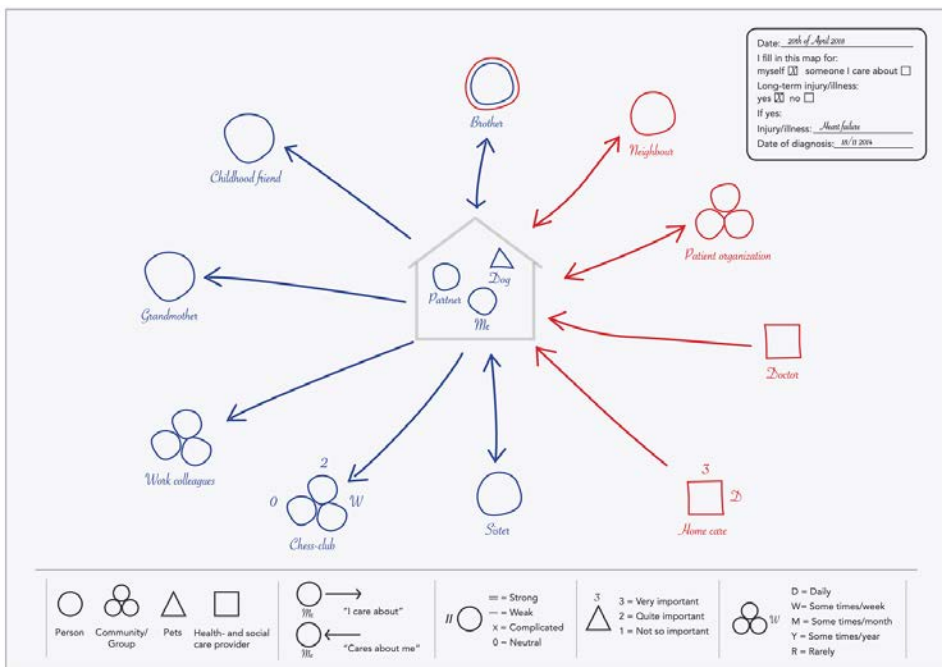
Figure 2. CareMaps illustration. Different social resources (e.g., persons, social groups, pets, and healthcare professionals) are represented by symbols. The type of relationship is indicated by using different colors (blue = social relations, red = supportive relations, groups, organizations, and healthcare). The direction of relationships (who cares for whom) is visualized using arrows. Finally, the quality of relationships is valued by adding symbols indicating the strength of the relationship (i.e., strong, weak, complicated, or neutral), its importance (i.e., not so important, quite important, or very important), and the frequency of interaction (i.e., daily, weekly, monthly yearly, or rarely). Reprinted with permission from (131). CC BY 4.0.

### 4.1.3 Study I

Study I explored types of social and healthcare relations that are important in the management of brain tumors (with varying tumor types, severity, and symptoms), as experienced by persons living with brain tumors, informal caregivers, and bereaved caregivers. The aim was to explore how persons living with brain tumors and informal caregivers perceive the potential usefulness of the CareMaps tool in their self-care and to describe the qualities of the interpersonal relations that they map.

#### 4.1.3.1 Study design

Study I was a qualitative study with semi-structured interviews. The interview guide comprised two main topics: impressions of the CareMaps tool and its potential usefulness in individuals' self-care and informal care, and experiences of supportive interpersonal relations and resources.

### 4.1.3.2 Study sampling

Participants were recruited in collaboration with the Swedish Brain Tumor Association, who organized workshops to which persons living with brain tumors, informal caregivers, and bereaved caregivers were invited to participate. The purpose of the workshops was to introduce the CareMaps tool, explain how to use it, and discuss how to develop it further. The participants were informed about the study in conjunction with the workshop sessions and invited to participate. All adult (>18 years) persons living with brain tumors, informal caregivers, and bereaved caregivers, irrespective of age, gender, or time since diagnosis who expressed an interest were eligible to participate in the study. Seven persons living with brain tumors, six informal caregivers, and six bereaved caregivers participated.

### 4.1.3.3 Data collection

The data collection consisted of a total of 19 individual face-to-face interviews conducted between January and December 2019. The audio-recorded interviews lasted between 23 and 119 minutes (median 60 minutes). As a preparation for the interviews, the participants, who had already been introduced to the CareMaps tool during workshops, were asked to use the tool to map relationships that they perceived as valuable. The intention was to stimulate reflection prior to the interviews.

### 4.1.3.4 Data analysis

Data was analyzed following the principles of thematic analysis with an inductive, semantic, and data-driven approach, aiming to identify patterns of shared meaning in the data set (132). The analysis was conducted in a stepwise fashion according to the chosen method of analysis. Familiarization with the data was achieved by reading the transcribed interviews and making notes on emerging themes. Interesting meaning units were identified and labeled with descriptive codes. In order to analyze the two topics of the interview separately, data related to the perceived usefulness of the CareMaps tool and data related to qualities of interpersonal relations were separated for subsequent per-topic analysis. Codes were categorized and the subsequent process of identifying, refining, and naming sub-themes and themes was performed iteratively in collaboration among all authors.

### 4.1.4    Study II

In Study II, I wanted to gain a deeper understanding of how informal caregivers actually use various resources to balance their caregiving with other aspects of life and manage challenges related to caregiving. It has been highlighted that strategies focusing on individualized support for informal caregivers (e.g., therapy services to improve physical and mental well-being, caregiving respite and relaxation) are still insufficient (133-135). Thus, exploring how informal caregivers of persons living with brain tumors use their intrapersonal, interpersonal, and healthcare and community resources could inform the development of better support services.

#### 4.1.4.1    Study design

Study II was a qualitative study with semi-structured interviews. Informal caregivers with at least one year of caregiving experience were interviewed on topics concerning management of challenges related to caregiving and available or lacking resources to support caregiving and other aspects of life including self-care. Coping theory was used as an analytic lens to gain a deeper understanding of how informal caregivers manage challenges related to caregiving, and which resources may be helpful in supporting various coping responses.

#### 4.1.4.2    Study sampling

Participants were initially recruited from among informal caregivers who had participated in Study I. Additional participants were recruited in collaboration with the brain tumor flow at Karolinska University Hospital. Informal caregivers who accompanied care recipients to hospital visits were informed about the study by clinicians. Those who expressed an interest were provided with written information, and upon their approval were contacted by the first author to introduce the CareMaps tool and schedule an interview. Eligible participants were adults (> 18 years) with at least one year of caregiving experience, and Swedish speaking. Purposeful sampling was conducted to achieve variation in gender and caregiving experience.

#### 4.1.4.3    Data collection

The data collection consisted of 16 individual interviews conducted either in person, by telephone or on Zoom, between August and December 2020. The audio-recorded interviews lasted between 43 to 114 (median 56) minutes. Prior

to the interviews, the participants were asked to use an updated version of the CareMaps tool to reflect on resources that are important to them. The updated version of the tool had a different layout than the initial version that was used in Study I, which aimed to reduce cluttering.

### 4.1.4.4 Data analysis

Data was analyzed iteratively in two phases using content analysis with a conventional approach followed by a directed approach (136). In the first phase, data was coded inductively, and related codes were grouped into categories, which were grouped according to the types of resources used (intrapersonal, interpersonal, and healthcare and community resources). In the second phase, a coding scheme was developed based on the Brief COPE instrument (73). The coding scheme specified 14 coping responses, grouped into three types: problem-focused coping (i.e., active coping, planning, using instrumental support), which aims to change the terms of the situation causing stress; emotion-focused coping (i.e., positive reframing, acceptance, humor, religion, using emotional support), which aims to manage the emotional distress associated with the stressful situation; and a third type of coping that involves responses that may be less useful and have been described as maladaptive or dysfunctional (i.e., self-distraction, denial, venting, substance use, behavioral disengagement, self-blame). The categories from the first phase were revisited and deductively coded based on the coding scheme to create sub-categories, labeled according to coping responses.

## 4.2 Exploring how patient-driven innovations can contribute to self-care and informal care

Having used a patient-driven innovation to identify resources that matter to persons living with a long-term conditions and informal caregivers in the first two studies, triggered questions about how patient-driven innovations may function as supportive resources for self-care and informal care. The CareMaps tool was still in an early stage of development and had not yet been widely distributed and used in self-care or in collaboration with healthcare. Meanwhile, colleagues in my research group published a scoping review exploring the nature and extent of patient-driven innovations published in peer-reviewed journals between 2008 and 2020 (116). The review included 96 publications and showed a clearly increasing publication trend in the 2010s. Based on publications identified in this review, I had the opportunity to explore the unmet needs that patient-driven

innovations address and how such innovations may be an entry point for patient innovators to engage in research and research publications. Thus, in studies III and IV, I broadened my focus to explore the nature of patient-driven innovations published in research literature.

### 4.2.1 Study III

Building on publications included in the scoping review, the aim of Study III was to explore what has been reported in peer-reviewed journals about the objectives and outcomes of patient-driven innovations, as well as their context of use (self-care and healthcare).

#### 4.2.1.1 Study design

Study III was a manifest content analysis of the publications included in the scoping review. In this study, I collaborated with an interdisciplinary team. The team included a patient innovator who was involved in all phases of the research process.

#### 4.2.1.2 Study sampling, data collection, and data analysis

We performed a secondary qualitative analysis of the publications that had been included in the previously published scoping review (116). The publications were analyzed inductively and systematically with a low level of abstraction to stay close to the data (137). The analysis process included collective reading and analysis of all 96 publications for unmet needs addressed by patient-driven innovations. The publications included original research, short reports, protocols, reviews, letters to the editor or commentaries, published conference abstracts, editorials, and special sections dedicated to patients (116). However, only original research publications, based on empirical data, irrespective of study design, were analyzed for outcomes. Each publication was individually analyzed by two researchers who extracted text corresponding to the content areas of interest into an Excel spreadsheet. The extractions were compared and discussed to resolve uncertainties and conflicts. The extracted data constituted the unit of analysis, from which meaning units were categorized into descriptive codes. The categorization of codes was discussed iteratively among all co-authors and refined until we reached agreement about the level of abstraction and the labelling of the categories.

### 4.2.2 Study IV

Several of the articles analyzed in Study III were authored or co-authored by patient innovators. This raised questions about their motives for and experiences of publishing in peer-reviewed journals. By increasing our understanding of how patient innovators reason about contributing to research production, the scientific community may be better prepared to support or partner with patient innovators in research. Therefore, the aim of Study IV was to explore patient innovators' reasons for and experiences of authoring scientific publications about their innovations.

#### 4.2.2.1 Study design

Study IV was a qualitative study with semi-structured interviews. In this study, I again collaborated with an interdisciplinary team including a patient innovator who was involved in all phases of the research process. Fifteen international patient innovators from three continents who had published in peer-reviewed journals were interviewed regarding the innovation journey, reasons for publishing, and experiences of the research and publication process.

#### 4.2.2.2 Study sampling

Participants were identified through the scoping review on patient-driven innovations (116). Of the 37 patient innovators identified through the scoping review, 28 had identifiable contact information. Nine of them responded and consented to be interviewed. An additional six patient innovators were recruited through snowball sampling. Fifteen patient innovators who had 1) (co-)developed a health innovation based on their experience as a patient or informal caregiver, and 2) (co-)authored at least one publication about their innovation in a peer-reviewed journal, were included in the study. The participants had developed various types of innovations (digital platforms, mobile applications, social innovations, and technical devices) for various long-term conditions (e.g., autoimmune diseases, diabetes, neurological conditions, and rare diseases).

#### 4.2.2.3 Data collection

The data collection consisted of 15 individual semi-structured interviews conducted between June and October 2022. The audio-recorded interviews lasted 31-74 (mean 50) minutes.

Collected data were transcribed verbatim and analyzed according to the Framework Method (138). The choice of method was motivated by its clear and systematic process of organizing data into a highly structured analytical framework, thus facilitating analysis in our interdisciplinary research team. First, three interviews were open-coded. The codes were discussed among co-authors who also contributed to create an analytical framework of codes grouped into categories. After the framework was tested on three additional interviews, it was iteratively refined, resulting in 37 codes and 17 categories. All interviews were independently coded using the framework. In the next step, the data was charted into a matrix, summarizing interviews per participant (in rows) by categories (in columns) with verbatim words underlined. Finally, themes and sub-themes were discussed and refined iteratively based on the matrix and guided by the research objectives.

## 4.3 Ethical considerations

### 4.3.1 Ethical approval

Ethical approval was granted for Studies I, II and IV. Ethical approval for Study III was not required as this study did not involve any primary data collection.

### 4.3.2 Informed consent

Participants in Studies I, II and IV were given written information about the purpose, scope, and procedures of the studies. They were informed that their participation was voluntary and that they had the right to withdraw at any time without giving a reason. They were also informed about confidentiality and management of personal data and pseudonymized communication of data. After voluntarily signing up for participation, the participants were contacted by phone or email for scheduling of interviews. Here, the study was discussed further, and the participants were able to ask questions prior to the interview and before giving informed consent. All participants consented to participation in writing or orally.

In studies I and II, we had to be particularly mindful in our selection of study participants. Persons living with brain tumors may suffer from one or multiple impairments (cognitive, neuropsychological, or functional), which may affect their ability to give informed consent (139). Recruiting participants in collaboration with the Swedish Brain Tumor Association (Study I) and healthcare

staff (Study II) helped us in identifying individuals that would be appropriate to approach for study participation.

### 4.3.3 Privacy and confidentiality

Personal data in all the studies is pseudonymized in accordance with the EU General Data Protection Act (140). The data was shared only among the researchers that were involved in the study. All interview files from Studies I, II and IV were securely stored on a safe server in accordance with the standards of Karolinska Institutet.

### 4.3.4 Risks and benefits

There were no clinical interventions in any of the studies in this doctoral project. However, there was a possibility that participating in the interviews (Studies I, II and IV) could cause unease or pose a risk of integrity violations through any inconvenience that may be caused by being observed and sharing personal experiences and reflections (141). Nevertheless, the benefits of this research were expected to be greater than the risks for the participants. Although not all participants in the four studies expected to benefit from this research directly, many expressed a wish to contribute to the development of new tools or functions or to making life easier for other persons living with long-term condition and informal caregivers.

### 4.3.5 Conflicts of interest

This doctoral research project was financed by the Swedish Research Council for Health, Working Life and Welfare and the Swedish Research Council (grant numbers 2014-4238 and 2018–01472). There were no further sponsors or funders involved in the design or execution of the studies (i.e., recruitment, data collection and management, analysis, write-up, and submission for publication).

The potential risks of conflict of interest were continuously discussed. For example, on the issue of recruitment and data collection, we considered the collaboration with the Brain Tumor Association and the associated risk of biased results (142). In Study I, participants were recruited from workshops, which were organized by a representative of the Brain Tumor Association who was also the developer of the CareMaps tool. Patients and informal caregivers were invited to the workshop, in part based on their activity in the organization and their interest in proactive and innovative solutions. No one from the Brain Tumor Association was present during any of the interviews, nor took part in the analysis of the data

or write-up of the manuscript. The representative read the final version of the manuscript. The results themselves do not present any direct or indirect benefits to the Brain Tumor Association.

# 5 Findings

## 5.1 Study I

Study I explored how persons living with brain tumors, informal caregivers, and bereaved caregivers perceived the potential usefulness of the CareMaps tool in their self-care, and how they described the qualities of their interpersonal relations.

### 5.1.1 Perceived usefulness of the CareMaps tool

The analysis yielded three themes reflecting the perceived usefulness of the CareMaps tool: Potential areas of use, Capturing the complexity of social relations, and Timing for introduction of the CareMaps tool (Figure 3).

Focus area                     Themes

Perceived usefulness of the CareMaps tool

- Potential areas of use
- Capturing the complexity of social relations
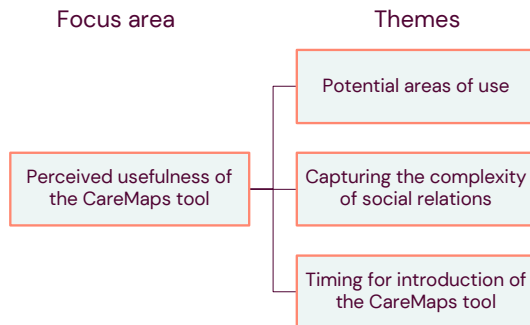- Timing for introduction of the CareMaps tool

Figure 3. Themes reflecting usefulness of the CareMaps tool as perceived by persons living with brain tumors, informal caregivers, and bereaved caregivers. Figure adapted from (131)

The findings show that participants' perceptions of the potential usefulness of the CareMaps tool were overall positive. In addition to providing an overview of the user's relations and resources, they found that it prompted self-reflection on one's own roles, attitudes, and approaches in regard to living with brain tumors or being an informal or bereaved caregiver.

> *Well, the first impression was … you don't understand how many relations you have. And there are many relations that perhaps you didn't think of and how those have changed. So that can be a lesson learned in itself. Also … you can be angry … because you think people have failed you or … so when you think about it, maybe that's not really it. Because you've been so caught up with yourself … maybe you don't even let people in. So, I think it is really important to sit down and reflect on your relations a little. (Bereaved caregiver 4)*

Participants highlighted that the illness situation, support needs, and the status of social relationships could change over time. With this in mind, participants shared suggestions for further development of the tool. These included extending the tool to capture the complexity and dynamics of social relations over time, including trust and sense of security. Participants also reflected on the potential use of the CareMaps tool for guidance and coordination of healthcare resources; and to include specifications of use at different time points and in different contexts (e.g., in self-care or in collaboration with healthcare).

### 5.1.2    Qualities in interpersonal relations

Two main themes were generated describing the types of relational qualities that the participants perceived as valuable: *Self-care supportive relations* and *Identity-preserving relations* (Figure 4).

Themes                                Sub-themes

Self-care supportive relations
- Family and friends providing committed and proactive support
- Persons with similar experiences having the ability to relate
- Healthcare staff using a person-centered approach

Identity-preserving relations
- Workplace supportive of professional identity
- Activities and contexts supportive of personal identity
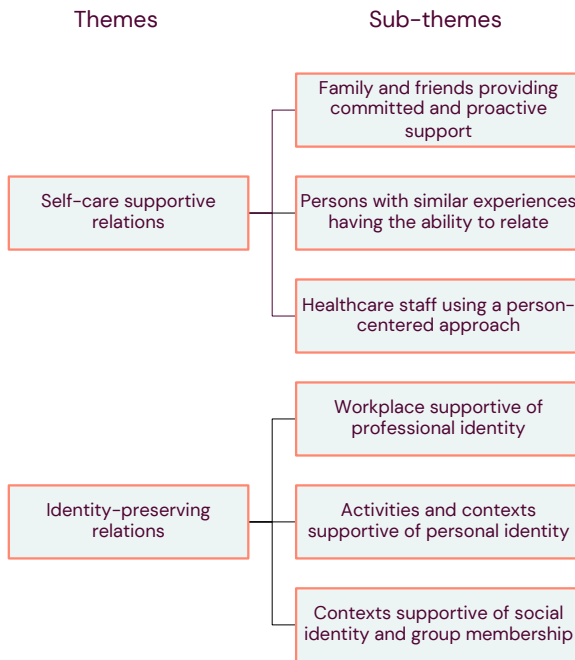- Contexts supportive of social identity and group membership

Figure 4. Themes and sub-themes reflecting qualities in interpersonal relations that matter to persons living with brain tumors, informal caregivers, and bereaved caregivers. Figure adapted from (131).

Self-care supportive relations were described as enhancing participants' ability to manage health and well-being in their illness-related roles. Such relations were found among participants' families, friends, and other well-established and trusted relationships providing committed and proactive support, and among persons with similar experience. Self-care supportive relations were also found

among continuous relationships with healthcare staff using a person–centered approach. These individuals were described as empathetic, and as acknowledging and respecting the experience and competence of the person living with brain tumors and of informal caregivers. Contact nurses in particular were described as an important resource for support and guidance for informal caregivers.

> *Their role in cancer care can't be stressed enough … they helped us through so many of those steps … [and] they gave an immense sense of security. We could call and they gave us answers, they asked a bit about how you were doing, gave you the warm pat on the shoulder. It became a little bit of a family…. Otherwise, you are pretty much left on your own. (Informal caregiver 4)*

Identity–preserving relationships and environments enabled the persons living with brain tumors and informal caregivers to avoid being defined by the illness or caregiving. Such relationships were found among both close social relationships and in wider social networks outside healthcare. Also, supportive colleagues that enabled participants to adjust their employment to fit their current capabilities and to facilitate absence during hospital visits, treatment periods, or temporary sick leaves were valued. The workplace also symbolized "business as usual" and allowed participants to focus on their professional competencies and expectations on job performance.

> *My job is extremely important. I can actually make a difference there, actually do something about things, unlike with mom's illness, where I can't do anything…. And the understanding [from the employer] is important. (Informal caregiver 5)*

Activities and contexts supportive of personal identity were described as important for persons living with brain tumors and informal caregivers. Meaningful social and physical activities and contexts contributed to a sense of normality. Social activities that may have become cognitively challenging were adjusted in time and scale rather than abandoned. Physical activities helped persons living with brain tumors to rehabilitate, keep fit, and maintain social contact, and the informal caregivers stressed physical activities as a source of recreation and respite.

## 5.2  Study II

In Study II, informal caregivers of persons living with brain tumors were interviewed about how they use intrapersonal and interpersonal resources to balance caregiving with other aspects of life and to manage challenges related to

caregiving. The analysis resulted in nine categories representing strategies that informal caregivers used to maintain a healthy balance between caregiving and other aspects of life (i.e., balancing strategies). Further, we identified seven coping responses that informal caregivers used to manage challenges. Figure 5 presents the findings organized by types of resources used.

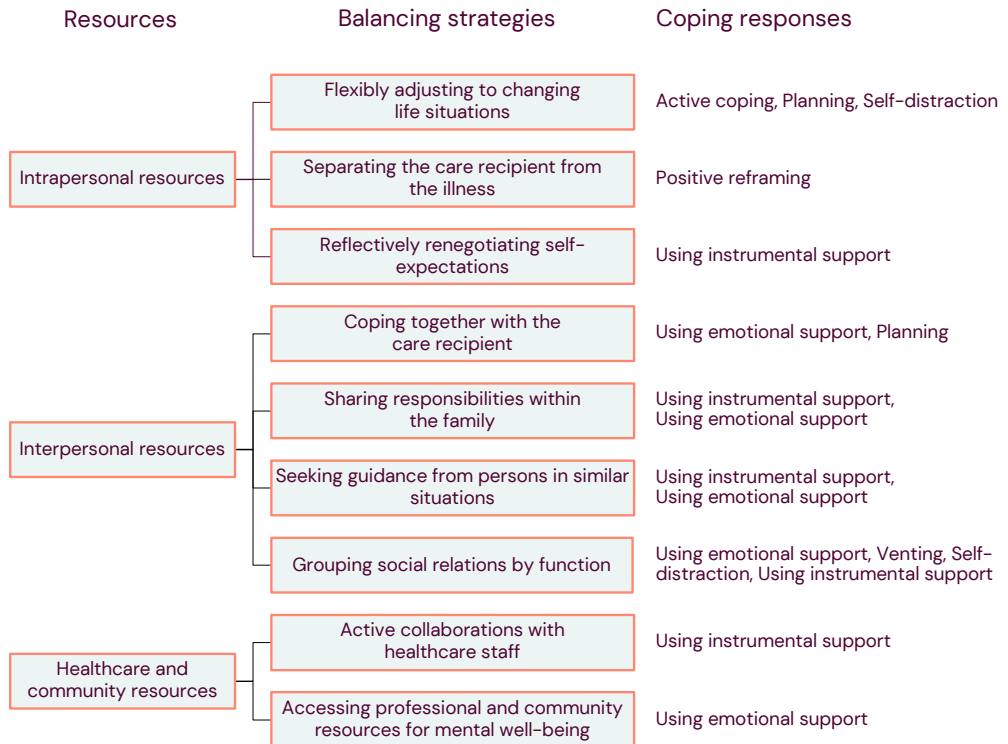| Resources | Balancing strategies | Coping responses |
|---|---|---|
| | Flexibly adjusting to changing life situations | Active coping, Planning, Self-distraction |
| Intrapersonal resources | Separating the care recipient from the illness | Positive reframing |
| | Reflectively renegotiating self-expectations | Using instrumental support |
| | Coping together with the care recipient | Using emotional support, Planning |
| Interpersonal resources | Sharing responsibilities within the family | Using instrumental support, Using emotional support |
| | Seeking guidance from persons in similar situations | Using instrumental support, Using emotional support |
| | Grouping social relations by function | Using emotional support, Venting, Self-distraction, Using instrumental support |
| Healthcare and community resources | Active collaborations with healthcare staff | Using instrumental support |
| | Accessing professional and community resources for mental well-being | Using emotional support |

Figure 5. Types of resources, balancing strategies, and coping responses used for maintaining a healthy balance between caregiving and other aspects of life.

### 5.2.1    Informal caregivers' use of intrapersonal resources

Informal caregivers described a changing illness situation that required continuous problem-solving, adjustments, and adaptations. Their coping responses included *planning*, *active coping*, and *self-distraction* to be able to balance caregiving with social and physical activities and make room for respite. Informal caregivers combined their gained experiences with *positive reframing*, enabling them to mentally separate the care recipient from the illness and symptoms (e.g., mood changes), and focus on their qualities, capabilities, and interests. To balance caregiving with other aspects of life, informal caregivers also renegotiated their self-expectations. Always putting the care recipient first

was described as physically and emotionally untenable, which led informal caregivers to reflect on their roles and self-imposed demands and make efforts towards asking for and *using instrumental support,* including paid household services.

> *I've become more egoistical. I don't have it in me, so I've had to work hard to put myself first … I've started to focus more on the little things like maybe to not rush home. Simple things like if I feel like having a coffee at a café in the sun. I wouldn't have done that before, because I got stuck in a notion that they need me more than they do. (Informal caregiver 1)*

### 5.2.2 Informal caregivers' use of interpersonal resources

Informal caregivers used their interpersonal resources for *instrumental* and *emotional support*. Family members, including the care recipient, were the nearest and most accessible social resources. Informal caregivers and family members had developed strategies for mutual emotional support, and practical sharing of household and caregiving responsibilities. Persons with similar experiences were also important resources of emotional and instrumental support; they were able to relate to experiences of illness and could provide illness- and healthcare-related information and guidance. Peer organizations or forums provided a platform to exchange experiences and support, as did persons with similar experiences in participants' existing social networks (e.g., among friends, relatives, or colleagues).

> *I have a friend who also … he doesn't have cancer, but a permanent illness. And so, I talk a lot with him … and he gives me perspective on what it's like being in my wife's situation. I mean, he's the equivalent … towards his wife … he's the sick one, so what's it like for him? And he has been ill for about as long [as my wife], and he also has children, and it affects his relationships a lot. (Informal caregiver 15)*

Close friends that informal caregivers described as meaningful and reliable social relationships were valuable supportive resources that allowed for unfiltered *venting* of emotions. Such friends also provided informal caregivers with opportunities for *self-distraction* and a mental or physical escape from the situation, which gave room for respite. The workplace provided a similar escape. Corroborating the findings of Study I, the participants experienced that focusing on work performance gave them a sense of control and normality. Supportive employers and colleagues facilitated combining work and caregiving and thus provided a meaningful environment separated from the illness situation.

### 5.2.3   Informal caregivers' use of healthcare and community resources

Informal caregivers reported limited or no healthcare support targeting caregivers specifically. This was described as frustrating by some, while others felt that the focus was rightfully on the care recipient. Informal caregivers received *informational* and *instrumental support* regarding the care recipients' clinical status and healthcare plans, but there was uncertainty regarding where to turn for additional support and guidance. In cases of collaboration and shared decision-making with healthcare staff, informal caregivers described an increased sense of inclusion. Contact nurses and so-called "neuro teams" were particularly mentioned.

> *When I told the contact nurse that [the care recipient's veins] had become difficult to puncture … they suggested to coordinate [treatment injections] and sampling … And the doctor's like "that's super smart" … And so, we decided that during the meeting. They have different roles and different perspectives on things, so that is perfect … Both to see that they respect each other's roles, but also that they have an open dialogue towards us and each other, is super important to our sense of security. Absolutely.*
> *(Informal caregiver 5)*

Some informal caregivers sought *emotional support* from professional support services (e.g., counselling, psychoanalysis) for themselves and for their family members. These services, for which they paid out of their own pocket, did not provide illness-specific support but provided important complements to healthcare and informal support resources.

## 5.3   Study III

Study III explored the objectives and outcomes of patient-driven innovations that have been published in the scientific literature, focusing on the unmet needs that such innovations address, and the reported outcomes for patients and healthcare. Of the 96 publications analyzed, thirteen were excluded as they did not include any information relevant to the study objectives. Thus, the analysis included 83 publications covering 21 patient-driven innovations, most of which were developed for use on an individual or community level without healthcare involvement. Reported outcomes of patient innovations based on empirical data were identified in 18 (22%) of the publications.

### 5.3.1   Unmet needs addressed by patient-driven innovations

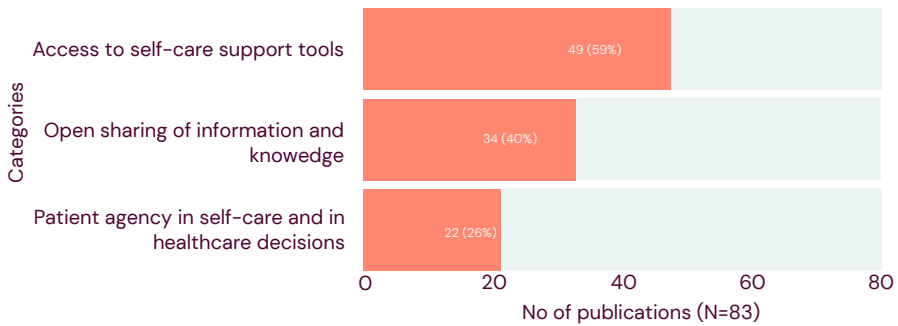We created three categories of unmet needs that the innovations addressed (Figure 6).

Figure 6. Number and proportion of publications represented in the three inductively created categories representing unmet needs addressed by patient-driven innovations. NB: some publications were included in more than one category. Figure adapted from (143).

### 5.3.1.1    Access to self-care support tools

More than half of the publications reported self-care support innovations developed for physical and social activities. These innovations often supported self-care monitoring to practically facilitate day-to-day life. For example, continuous monitoring for treatment follow-up and sharing of health data (144), or monitoring and preventing stoma bag leakage using a sensor developed from existing technology (145). Other innovations for self-care support comprised non-technological tools and products for physical functioning and access to participation in social activities (146, 147). Some innovations consisted of creative ideas for using existing tools or technology and involved little or no product development (148).

### 5.3.1.2    Open sharing of information and knowledge

More than one third of the publications concerned platforms for open sharing of self-care-related information and knowledge. These innovations included platforms that enabled creation, sharing, and comparing of personal health data profiles for various health conditions (149, 150). Such platforms serve to enable people to compare and discuss issues ranging from individual symptoms and treatment history to lived experiences and self-care routines, to empower people to make informed care decisions (151). Other patient innovators hosted platforms for open sharing of code and provision of assistance for patients who wanted to build do-it-yourself solutions (152). Making do-it-yourself technologies more broadly available enabled patient collaborations and accelerated development and access of innovations.

## 5.3.1.3   Patient agency in self-care and healthcare decisions

About one quarter of the publications concerned patient-driven innovations addressing the unmet need of patient agency in self-care. These innovations aimed to provide trustworthy information and knowledge through shared libraries of quality-assured diagnosis-specific self-care information (153), personal health records (154), and online health information platforms (155) for collecting, sharing, and comparing health data over time, and with peers.

Other patient-driven innovations aimed to improve collaboration with healthcare. For example, mobile patient support systems enabling direct communication and information exchange between patients, families, and their care teams (156), or enabling integration of patient-reported data into the patients' electronic medical record (157). Innovations also included the establishment of collaborative multi-professional networks including patients and informal caregivers to increase patient agency in strategic healthcare decisions (e.g., the design of care pathways) (158).

## 5.3.2   Reported outcomes of patient-driven innovations

We created two categories reflecting reported outcomes of patient-driven innovations (Figure 7).
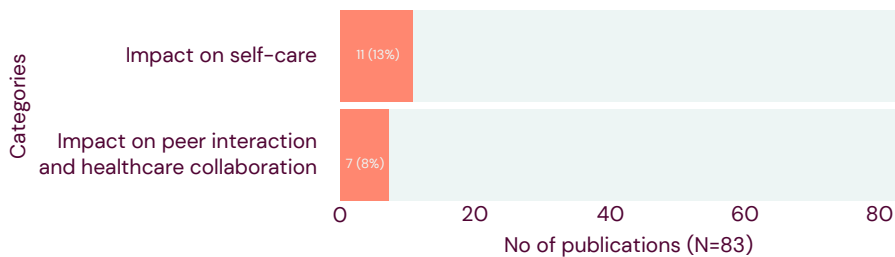


Figure 7. Number and proportion of publications in the two inductively created categories representing outcomes of patient-driven innovations. NB: in total 65 (78%) of the publications did not report any outcomes. Figure adapted from (143).

## 5.3.2.1   Impact on self-care

Eleven publications reported impacts on self-care; these included perceptions of improved self-care processes; health outcomes (e.g., improvements in health parameters); and well-being (e.g., overall reduced burden of illness). Several of the innovations that were evaluated with positive outcomes were so called do-it-yourself technologies for diabetes type 1. Although they expressed uncertainty and concerns about such technologies, healthcare professionals were open to

participating in educational training. Positive impacts on self-care were also reported for other types of innovations. For example, an innovation for gait training improved functional motor skills among children with cerebral palsy (147, 159); and a jacket with pockets for drainage tubes for women with breast cancer increased their social interaction (146).

### 5.3.2.2  Impact on peer interaction and healthcare collaboration

Five of seven publications that reported outcomes related to peer interaction and healthcare collaboration were based on one innovation (*PatientsLikeMe*) (160). One publication highlighted the potential challenges in online patient communication due to variations in use of terminology when describing symptoms. Two publications reported strengthened patient agency in collaborations with healthcare through platforms and support systems, enabling active engagement of both patients and healthcare professionals, and improved communication. None of the publications reported any negative outcomes.

## 5.4  Study IV

Study IV explored patient innovators' reasons for and experiences of authoring scientific publications about their innovations. The analysis resulted in three themes reflecting patient innovators' reasons for publishing and four themes reflecting experiences of publishing (Figure 8).
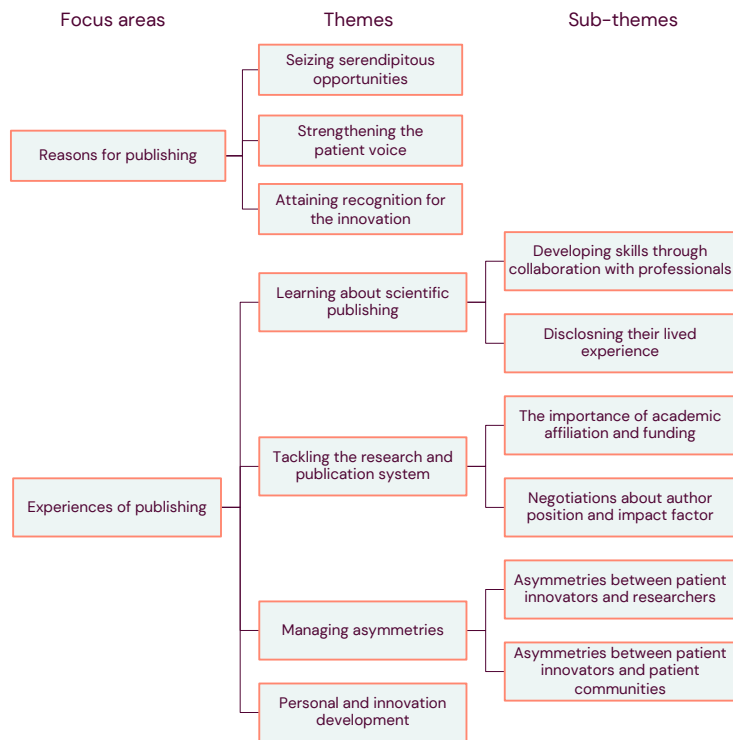
Figure 8. Themes and sub-themes reflecting patient innovators' reasons and experiences of authoring scientific publications about their innovations.

### 5.4.1 Reasons for publishing

The findings of Study IV show that in addition to developing innovations to support self-care, patient innovators aspired to increase the meaning and relevance of research and development of healthcare services for persons living with long-term conditions and informal caregivers. Some participants described that at first, they may not have understood the benefits of publishing. The decision to publish was more about being in the right place at the right time rather than an ambition. Once published, some became motivated to publish again. One reason given for engaging in scientific publishing was to strengthen the patient voice. Participants emphasized the importance of effecting a culture change by bringing attention to specific patient communities (e.g., rare diseases), sharing self-collected data to increase real-world evidence in research, and increasing collaborations with equitable involvement of patients and informal caregivers throughout the entire research process. Another reason was to attain recognition for the innovation within the healthcare and research communities, and to contribute with their lived experiences and with their

innovation competence to improve research and health outcomes. Although not their primary communication channel, patient innovators considered scientific publishing to be the most effective way to gain attention and recognition among key stakeholders such as healthcare and academia.

> *Publications are the space where [scientists and companies] get their information. So it was really important to be able to reach people where they are … sort of entering their world … where they learn, and they gather information. (Patient innovtor 11)*

### 5.4.2   Experiences of publishing

#### 5.4.2.1   Learning about scientific publishing

Participants described that collaborations with supportive researchers had enabled development in the patient innovators' research and writing skills, which helped them to achieve their main priority – to convey the patient perspective. Whether to disclose their patient or informal caregiver status depended on the purpose of the publication; they would disclose their status when sharing their lived experiences, but not when reporting on epidemiological or clinical research. Another reason for not disclosing their status was their experience of mixed reactions (either appreciation or questioning their credibility as scientific authors) from the scientific community.

#### 5.4.2.2   Tackling the research and publication system

Participants experienced obstacles in the research and publication system. They stressed an urgent need for "democratizing research" to remove barriers and facilitate people outside the research community to publish and access published data for free. Participants also questioned the long publication process, which they perceived was in part due to negotiations about journal impact factors (i.e., measure of the annual citation frequency of journals) and authorship position (i.e., order of appearance among the list of authors of a publication).

> *It took a long time to get it published, because in the beginning everybody wanted to be number one and four … and then, you know, to find the right journal and then to go through the process we have to keep redoing … and this and that. They put us through the mill. I can't believe it actually got published.*
> *(Patient innovator 3)*

### 5.4.2.3   Managing asymmetries

Patient innovators perceived that their lived experience was an important complement to researchers' scientific knowledge and skills. Some experienced positive collaborations defined by transparency, mutual respect, and active participation in integrating the two perspectives.

> *[The research team] was really open and willing to integrate our point of view.… What has worked well is with the way we were able to provide our feedback and provide our expertise [and] experience within the publication. The fact that we were encouraged and supported to be part of the scientific process … I'm thinking about publishing again. (Patient innovator 13)*

Negative experiences of perceived asymmetries were also reported. Patient innovators' lack of research training or a publication record was a barrier to being acknowledged by the scientific community. Also, living with long-term conditions or providing informal care, having another professional occupation, and limited or no research funding and resources offered by a university (e.g., coverage of open access publishing fees or access to publications behind paywalls) were perceived as asymmetries between patient innovators and researchers. Such barriers were described as hindering patient innovators from full-time engagement in research and publication processes.

Participants also reported asymmetries between themselves and the patient communities that they represented. They believed that their experiences of advocating for the patient voice distinguished them from the majority of patients and informal caregivers, and therefore they felt an obligation to represent the patient and informal caregiver communities they identified with.

> *I think that within patient-led innovation or patient [involvement in] publication we need to make sure that [we] reflect not only the voice of the educated ones and the knowledgeable ones, but also reflect the voice of the ones who do not have a voice. (Patient innovator 13)*

### 5.4.2.4   Personal and innovation development

Participants experienced that their learning experiences from research and publication (e.g., regarding methodology and data collection) had advanced the development of their innovations. In addition, their experiences of research publication processes had resulted in scientific evidence which enabled more research and additional publications, which in turn, for some, had led to new collaborations.

# 6 Discussion

## 6.1 Main findings

This thesis contributes to an increased understanding of supportive resources that matter to persons living with long-term conditions. Additionally, the thesis demonstrates how patient-driven innovations address unmet needs related to individuals' self-care and how patient innovators can contribute to research production. Studies I and II focused on supportive resources that matter to persons living with brain tumors and informal caregivers and explored the usefulness of a caregiver-developed tool to identify such resources; and Studies III and IV focused on driving forces for the development and dissemination of patient-driven innovations in a wider context, as well as outcomes of patient-driven innovations and patient innovators' experiences of publishing.

The first part of the discussion will focus on the findings from Studies I and II regarding informal caregivers' experiences and use of interpersonal resources. The second part of the discussion will focus on different types of patient-driven innovations and how they relate to the supportive resources for self-care and informal care that were identified in Studies I and II.

## 6.2 Discussion of the findings in relation to the literature

### 6.2.1 Supportive resources that matter

The CareMaps tool that was used in the first two studies helped respondents to identify supportive resources in their social and healthcare networks, as well as reflect on their own roles, attitudes, and experiences. I will begin by discussing intrapersonal resources that were identified as important in the two studies, followed by interpersonal and healthcare resources.

#### 6.2.1.1 Intrapersonal resources

The use of intrapersonal resources, and specifically coping strategies that informal caregivers used to balance caregiving with other aspects of life, was explored in Study II. We found that informal caregivers' ability to plan their time and activities was important for flexibly adjusting to their changing life situation. Informal caregivers emphasized the importance of focusing on other things than the illness. They could achieve this by planning activities that distracted them from the situation. In addition, positive reframing was used to separate the care

recipient from the illness. In the context of brain tumor care, it has been found that such positive thinking strategies may have a favorable impact on quality of life (75). These resources (i.e., the ability to plan, distract oneself, and think positively), which informal caregivers used to balance caregiving with other aspects of life and thus focus on their own self-care and wellbeing, suggest that caregivers were able to positively adapt to stressful situations and reduce caregiver burden (161, 162). However, these resources may not be available to everyone. For those suffering from high caregiver burden, balancing caregiving with other aspects of life may be particularly challenging. This was partly highlighted by another important intrapersonal resource that we identified, namely the ability to renegotiate self-expectations. In their narratives, informal caregivers described how they sometimes had to accept and rely on help from others (e.g., with cleaning or household duties). Our findings suggest that the need for support may not have been self-evident for informal caregivers. This is in line with previous research suggesting that informal caregivers may be hesitant to seek external support, which may be due to a belief that they need to rely on themselves, or experiences of stigma(163-165) . This emphasizes the importance of proactive outreach with support services to complement informal caregivers' own resources.

### 6.2.1.2 *Interpersonal resources*

The findings of Studies I and II show that the mapping and visualization of interpersonal relationships stimulated reflection about the value of social relationships, beyond quantity and structure. Visualizing interpersonal relationships enabled persons living with brain tumors and informal caregivers to identify the qualities (Study I) and areas of use (Study II) of supportive resources for individuals' self-care and informal care and to minimize the impact of illness on everyday life. In Study I, two themes were created to describe the types of relational qualities that the participants described as valuable: self-care supportive relations, which enhanced participants' ability to manage challenges and maintain health and well-being in their role as either a person living with long-term conditions or an informal caregiver; and identity-preserving relations, which enabled participants to avoid being defined by the illness and illness situation.

Self-care supportive relations were found within various social groups (e.g., family, friends, persons with similar experiences, and healthcare staff). Identifying with and belonging to different social groups has been shown to increase access

to social support and the likelihood of seeking social support (166-168). For example, persons with similar experiences were valued for their ability to relate to participants' situation and provided emotional support and a sense of belonging. In addition, persons with similar experiences had relevant knowledge and competencies and could provide informational support and guidance on navigating the healthcare system. Previous research has shown that persons with the ability to relate have a positive impact on people's willingness to give and receive support (169, 170). Some participants found persons with similar experiences among friends, relatives, or co-workers. Others commended peer organizations (e.g., the Brain Tumor Association) for facilitating peer gatherings that enabled new and meaningful relationships in an open yet safe environment for sharing of experiences.

In Study I, healthcare staff were included among self-care supportive relations. Relations with healthcare staff were characterized by qualities such as empathy, continuity, and inclusion. Healthcare staff, in particular contact nurses, using a person-centered approach were valued as supportive to persons living with brain tumors and informal caregivers. Previous studies suggest that nurses acting as a primary point of contact have a relational function and may constitute a resource to informal caregivers, enhancing their inclusion in the care team (171, 172). Interestingly, while the findings of Study I emphasized the emotional support that contact nurses could provide by showing empathy, the findings of Study II emphasize the instrumental support provided by healthcare staff in communicating individualized information regarding the care recipient's illness status and treatment. Both studies found that although there were positive experiences of support provided by healthcare, there were also experiences of a lack of inclusion in and collaboration with the healthcare team. Informal caregivers expressed a desire and need for inclusion in the healthcare team, which is in line with recommendations to improve their preparedness for daily caregiving (173).

Identity-preserving relations were valued for enabling membership in social groups or contexts disconnected from the illness experience. The respondents in both Studies I and II described that they did not want to identify themselves exclusively as persons living with an illness or informal caregivers. Rather, they expressed a need to identify themselves also according to other aspects of life, such as on a professional, personal, and social level. Having multiple social identities (e.g., that of an informal caregiver, a parent, and a co-worker) and multiple group

memberships that are perceived as positive has been shown to enhance well-being (168, 174).

Identity-preserving relations were found among close social relations, as well as in wider social networks. The findings in Study I showed that identity-preserving relations included social forums and physical activities that offered momentary respite from illness and caregiving. It has been found that some persons living with cancer and informal caregivers may strive for normality by distancing themselves from the illness (42). Disconnecting from the patient or informal caregiver identity may enable other identities (personal, social, or professional) and enhance sense of self (175). A supportive workplace and colleagues were emphasized as important for providing a sense of normality, which corroborates findings from previous research (176). The workplace was found valuable for participants' professional identities, and as a resource for self-distraction from the illness situation.

### 6.2.2    The roles of patient-driven innovations

The use of the CareMaps tool in the first two studies illustrated its role mainly as an instrument to support data collection and identify resources that matter to persons living with long-term conditions and informal caregivers. Although this thesis does not provide any empirical evidence of the usefulness of the CareMaps tool as a supportive resource in individuals' self-care and informal care, Study I provided some insight into how persons living with brain tumors and informal caregivers reasoned about potential uses of the tool. Respondents raised questions regarding the optimal timing for using the tool, and what kind of role the tool could play in supporting self-care, informal care, and collaboration with healthcare. These questions are relevant to explore further, in particular as support needs change during the trajectory of brain tumor illness (30, 177). Rather than focusing exclusively on the CareMaps innovation, I chose to broaden my focus to additional patient-driven innovations for which more empirical data are available. By exploring the objectives and outcomes of patient-driven innovations published in the research literature in Study III, I was able to identify various roles played by patient-driven innovations, namely as self-care support services, platforms for open sharing of information and knowledge, services for facilitating collaboration with healthcare, and multiprofessional networks for healthcare improvement initiatives. In the subsequent sections, I will discuss these roles in relation to the supportive resources identified in Studies I and II.

### 6.2.2.1    Self-care support services

The findings in Study III show that more than half of the patient-driven innovations identified through a scoping review were self-care support tools, addressing an unmet need for self-care supportive resources (116). These innovations involved services for self-care monitoring and tools for supporting physical functioning, which may complement self-care supportive relations. Further, we identified patient-driven innovations that aimed to facilitate participation in social activities. An example was a special jacket developed for breast cancer patients (*Jacki Jacket*) which contributed to making them feel more comfortable about their physical appearance (146). This was a patient-driven innovation that could support individuals in attaining or maintaining important social relationships and participate in social activities, which was emphasized as an identity-preserving resource in Study I. Adding to the importance of social networks, active participation in social activities has been shown to increase health-related quality of life (178). Although studies reporting on outcomes of patient-driven innovations are still limited, some of the reviewed publications reported on contributions to improved self-care processes, health outcomes, and well-being among end users (149, 159, 179).

### 6.2.2.2    Platforms for open sharing of information and knowledge

One third of the publications in Study III reported on platforms for open sharing of self-care related information and knowledge. These included social platforms to enable peer interaction (e.g., through joint chat forums) and sharing of personal health data and self-care recommendations (150, 180). These solutions illustrate how patient-driven innovations could facilitate the establishment of "peer relations", i.e., relations with persons who have similar experiences and were identified as self-care supportive resources (Study I), providing both instrumental and emotional support (Study II). Examples of instrumental support facilitated by platforms such as *PatientsLikeMe* included the exchange of questions and advice related to self-care (151, 181). It has been shown that individuals who use platforms for sharing of personal health data mainly do so to improve their own self-care, rather than focusing on helping others (182). This contrasts with the altruistic motives that have been reported to drive patient innovators to openly share do-it-yourself solutions to contribute to others' self-care (152).

### 6.2.2.3  Services for facilitating collaboration with healthcare

Some patient-driven innovations were developed to facilitate communication and partnerships with healthcare by enabling information exchange and health data integration (145, 156, 157). The *Genia* app, targeting persons living with cystic fibrosis and their family networks, is an example of how patient-driven innovations could facilitate the involvement of informal caregivers in collaborations with the healthcare team, which was emphasized as important among informal caregivers in Studies I and II (156). However, only a few publications reported on actual use or outcomes of patient-driven innovations in healthcare. The value of end-user innovations is often underestimated among decision-makers and policymakers, which could challenge the use of such innovations in healthcare (183, 184). To facilitate the process of regulatory approval and implementation of patient-driven innovations, it has been suggested that a multidisciplinary approach involving collaboration among patient innovators, healthcare staff, and researchers is needed (115, 185).

### 6.2.2.4  Multiprofessional networks for healthcare improvement initiatives

Some of the patient-driven innovations identified in Study III were not tangible tools or services but could be described as new approaches to addressing healthcare challenges. We identified patient- and caregiver-driven collaborative multiprofessional networks that involved patients, informal caregivers, clinicians, and researchers (158, 186, 187). These networks had in common that various actors collaborated in the co-design of improved care services, representing examples of co-production in healthcare (188). These patient-driven innovations contributed to increasing patient and caregiver agency beyond the encounter with healthcare staff, allowing them to influence strategic decisions regarding the design of healthcare services. There is evidence from previous studies that co-creation with patients in the design and planning of care can have several benefits, such as improved patient-professional relationships, improved health outcomes, and increased satisfaction among clinicians (189). Co-creation processes involving patients in healthcare improvement work have received much attention in recent years, in particular using methods such as experience-based co-design (190, 191). However, less attention has been devoted to exploring patient-driven co-creation processes. We found that patient innovators co-authored some of the publications included in Study III, which provided a base for exploring another role of patient-driven innovations as an entry point to research collaborations.

### 6.2.3 Patient innovators' reasons and experiences of scientific publishing

The findings of Study IV show that patient innovators engaged in scientific publishing about their innovations to strengthen the patient voice and attain recognition for their innovations. Some were invited to participate in research collaborations as a consequence of presenting their innovations at conferences, which indicates that their contributions were valued by the research community. Thus, their innovations served as an entry point to planned or unplanned research collaborations. Previous research has highlighted various benefits of involving persons with lived experience in research collaborations, such as contributing to more relevant research questions and study designs, as well as the interpretation of data and communication of findings in accessible language to end users (192, 193). Patient innovators can contribute with additional competencies and experiences acquired through developing and disseminating innovative solutions, tools, and services for patients and informal caregivers.

The patient innovators in Study IV emphasized that the support of researchers was key to their engagement in research. They experienced that successful research collaborations were defined by mutual respect and trust, sharing of power, and recognition of the competence and knowledge of all stakeholders involved. This supports conclusions from previous research that building research partnerships involving persons living with long-term conditions takes time and demands a focus on the partnership process rather than the outcomes (194). A study on the co-creative research process in the "Patients in the driver's seat" program showed that it can take years for members to view one another as individuals with individual competencies, rather than simply grouping members into their roles as 'patient innovators' or 'researchers' (195). The findings of Study IV suggest that consistent and continuous research collaborations where differences are recognized as assets can facilitate publications as well as the development of innovations, ultimately supporting individuals in their self-care and informal care.

## 6.3 Methodological considerations

Qualitative analysis was used in all four studies. The choice of design was driven by the character of the research questions. Qualitative research is suitable for exploring individuals' experiences and perspectives of a phenomenon (196). A descriptive approach was used to increase understanding of patient-driven innovations, and of participants' reasoning and experiences in relation to them,

from both a developer and an end-user perspective, while keeping a low level of interpretation (197).

### 6.3.1 Selection of participants

In Studies I and II, being Swedish speaking was a criterion for participation. This allowed for rich and nuanced narratives and minimized language barriers and the risk of miscommunication and misinterpretation. However, it may have excluded potential participants and thus reduced the variation of experiences shared (198). The gender distribution in Study I was not representative for the population. The majority of persons living with brain tumors were men (5 of 7, 71%) and almost all informal caregivers (11 of 12, 92%) were women. According to estimates, gender distribution is fairly equal among persons living with brain tumors and among the population providing informal care in Sweden (36, 126). In Study II, the gender distribution of informal caregivers (38% men, 62% women) was more balanced. It should also be considered that there are gender differences between informal caregivers. For example, women are often more affected by caregiver burden and it is more common among women than men to reduce work hours or to take other measures leading to lower income and pension (36, 199). In addition, women experience a higher physical and psychological caregiver burden, and more difficulties combining caregiving with other leisure activities (46, 200). Therefore, an inclusion of more male caregivers may have added other perspectives to the research questions in Studies I and II.

### 6.3.2 Selection bias

There was a risk of selection bias towards engaged and proactive individuals with the ability and capacity to attend peer organization gatherings in both Studies I and II (201). In Study IV, all identified patient innovators who had authored publications included in the scoping review (N=37) were approached. Although the number of patient innovators who have published about their innovations is increasing, it is still low, which may have impacted the variation of characteristics among the participants. In addition, author roles are not always well-described in scientific articles, which may make it difficult to identify patient innovators as co-authors, and consequently, some patient innovators that had published may have been excluded (116, 202).

### 6.3.3 Information power

The concept of information power has been considered throughout the studies of this thesis. Information power refers to the capacity of a sample to provide information that is rich, meaningful, and relevant to the research question (203). Information power depends on whether the *aim* is narrow or broad, if the *sample specificity* is dense or sparse, whether there is a *use of theory*, strong or weak *quality of dialogue*, and case or cross–case *analysis strategy*. We have striven for high information power through narrow aims, a focus on the specific experiences, knowledge and competences of participants, focused interview dialogues based on guides, and analysis conducted both across and within data sets.

### 6.3.4 Trustworthiness

Trustworthiness concerns the evaluation of procedures used to generate the findings of qualitative studies (137). Credibility, how well the data process and analysis match the research question; dependability, the degree to which data may change over time and the consistency of data collection; and transferability, to what extent the findings can be transferred to other settings, are all aspects of trustworthiness.

#### 6.3.4.1  Credibility

In Study I, I chose to use data–driven thematic analysis with a semantic approach, following the six steps proposed by Braun and Clarke (132). The choice of analysis method was suitable to provide rich and detailed descriptions of the data, regarding both perceptions of use and reasoning about the qualities of interpersonal support resources (132). In Study II, we conducted a two–step content analysis with both a conventional and a directed approach guided by coping theory (73, 136). The two–step analysis enabled an inductive immersion and categorization of the data, and the application of coping theory on the inductive categories to link informal caregivers' use of various resources to coping responses. In Study III, we used manifest content analysis to describe the data characteristics by systematically coding the data and staying close to the text throughout the analysis process. The choice of analysis method was appropriate as we wanted to keep a low level of interpretation and abstraction when identifying the unmet needs addressed by patient innovations and the reported outcomes from using them (137, 204). In Study IV, data were analyzed using the Framework Method (138). This systematic method of organizing data was selected to facilitate the analysis process in our interdisciplinary research

team. The choice of analysis method supported the aim to explore experiences of scientific publishing among patient innovators by identifying commonalities, differences, and relationships among the data. The use of quotations in Studies I, II and IV helped to illustrate the particularities of the categories or themes in relation to each other and thus enhanced credibility.

### 6.3.4.2 Dependability

Dependability concerns stability of data and to what degree results may change over time (205). It may be difficult to obtain in qualitative research given the limited possibilities of replicating studies in the same context, using the same methods on the same participants (206). However, attempts were made through discussions among co-authors, and transparent and detailed methodological descriptions of the research design and implementation, mode of data collection, and mode of analysis to allow for the study to be repeated.

### 6.3.4.3 Transferability

Transferability is the extent to which the findings can be applied to other settings or groups beyond the study setting (196). Studies I and II were conducted in Sweden among persons living with brain tumors, informal caregivers, and bereaved caregivers about their perceptions and experiences of supportive resources in self-care and informal care. In both studies, the qualitative analysis resulted in generic categories without specific references to living with brain tumors. The generic nature of the resources valued for self-care and informal care enhances the chances of transferability to similar contexts. However, in other settings, additional resources may be identified as important in supporting self-care and informal care. Studies III and IV were conducted in an international setting of various types of patient-driven innovations originating from three different continents, developed for various long-term conditions and published in scientific journals. Whether the findings are transferable to patient authors who do not identify as patient innovators, or to patient innovators without experience of scientific publishing, would merit further investigation. All studies report thorough descriptions of the study contexts, characteristics of selected participants, and data collection and analysis processes to help readers in determining the transferability to similar contexts (207).

### 6.3.4.4 Reflexivity

Reflexivity concerns the ways in which the researcher's background, position, and perspectives affect their ability to remain neutral in the research process (196). My personal experiences of informal cancer caregiving and bereavement, and of the impact of social relationships and support on coping, gave me a preunderstanding that enabled me to identify with many of the experiences shared by the participants, especially in Studies I and II. Meanwhile, these experiences have obliged me to be self-aware and to critically reflect on my own subjectivity and preconceptions and their possible impact on my role as a researcher (196). Efforts to prevent "going native" (208) and to maintain neutrality were made through discussions about the study designs, the interview guides, and analysis processes in the interdisciplinary supervisor group as well as with co-authors, which also ensured confirmability.

### 6.3.4.5 Reciprocity

Reciprocity concerns the importance of mutual exchange, respect, and transparency in the researcher-participant relationship (209). It recognizes that participants contribute to the study with their time, experiences, knowledge, and insights. Participation in Studies I, II and IV was voluntary and, as expressed by participants, of interest and importance. However, it depends on active participation and sharing of personal experiences and reflections. The participants were reminded that they had the opportunity to break or stop the interviews at any time. All the interviews were conducted at a place (digital or physical) and time most convenient for the participants, regardless of time zone. The findings were communicated via e-mail to the participants after the studies had been completed.

# 7 Conclusions

This thesis contributes to knowledge about supportive resources that matter to persons living with long-term conditions and informal caregivers, and how patient-driven innovations can facilitate self-care and informal care.

Our findings indicate that membership in various social groups and contexts was perceived as important for self-care and informal care among persons living with brain tumors, informal caregivers, and bereaved caregivers in Sweden. Self-care supportive relations were identified as particularly important for managing uncertainties and challenges related to self-care and informal care. In contrast, identity-preserving relations were important for enabling membership in social groups and contexts disconnected from the illness. Informal caregivers used a combination of intrapersonal, interpersonal, and healthcare and community resources to balance caregiving with other aspects of life and manage challenges related to caregiving. Although these findings are limited to the context of brain tumor care in Sweden, similar resources may be important to persons living with long-term conditions and informal caregivers in other contexts.

The role of patient-driven innovations was explored in and beyond the context of brain tumor care. The CareMaps tool was found to be useful for stimulating self-reflection about important supportive resources, and specifically the quality of social relationships, among persons living with brain tumors and informal caregivers. Based on published research literature, we found that patient-driven innovations play various roles in supporting self-care, collaboration with peers, and collaboration with healthcare, and in establishing research partnerships. Although reported outcomes of patient-driven innovations are still limited, our findings suggest that such innovations can act as self-care supportive resources and facilitate the establishment of relationships that matter to persons living with long-term conditions and informal caregivers.

All studies in this thesis illustrate that persons living with long-term conditions and informal caregivers are central stakeholders in driving healthcare development and research. Patient innovators shared their accounts of how they contributed with their lived experience and innovations to strengthen the patient voice in research. Despite positive experiences of research and publication processes, patient innovators faced cultural and structural barriers. Our findings elucidate that continued efforts are needed to facilitate for patient innovators,

as well as other patient and public contributors, to contribute with their experience and expertise to the production of relevant and meaningful research and services supporting self–care and informal care.

# 8  Points of perspectives

The findings from this thesis have implications for practice and future research. Patient-driven innovations are gaining ground, while research on the outcomes of such innovations is still limited. More empirical evidence is needed to demonstrate the value of patient-driven innovations as supportive resources for self-care and informal care. In particular, there is limited evidence of the use of patient-driven innovations in healthcare. The CareMaps tool is an example of a caregiver-developed innovation that proved valuable in identifying existing and potentially missing resources in individuals' self-care and informal care. Future research should further investigate how the CareMaps tool can be used to facilitate self-care and informal care for persons living with various long-term conditions. Further, research may address how the tool could be used in collaboration with healthcare staff to help persons living with long-term conditions and informal caregivers to communicate their existing resources and support needs to healthcare staff.

Our findings indicate that persons living with brain tumors and informal caregivers valued membership in various social groups and contexts within and outside their closest social networks. Future studies may explore how well the qualities identified in those relationships fit with other populations and contexts. In addition, there is a need for further investigation of how social network-mapping tools that capture quality in social relations should be designed, and how to develop interventions that provide appropriate support for individuals' self-care and informal care, while protecting their professional, personal, and social identities.

Multiprofessional collaborations between various stakeholders, involving patient innovators, healthcare professionals, researchers, and policymakers, will be needed in order to perform evaluation studies and support the future adoption of effective innovations in various contexts, including healthcare. This thesis reports examples of successful collaborations between patient innovators and researchers, barriers to patient and public involvement remain. Therefore, we recommend that future research should explore how to build supportive structures for multidisciplinary collaborations that can contribute to quality assurance and evaluation of patient-driven innovations.

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