Patient and next-of-kin collaboration for better research and healthcare

COLLABORATION 2.0: SUSTAINABLE COLLABORATION FOR VALUE AND INNOVATION

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Summary

In this report, we have built on established frameworks and toolkits and drawn on the documented experiences of collaboration between patients, next of kin, research and healthcare. Collaboration in this area often refers to a way of addressing challenges through representation from different types of stakeholders.

It is clear from the reviewed material that there is a strong desire for collaboration, but that many people make similar mistakes. For example, setting clear objectives for collaboration from the outset or defining evaluation criteria are things that are often forgotten.

These challenges contribute to the misconceptions and ambiguities surrounding collaboration. Many people want to collaborate, but what does collaboration lead to? How do you do it in a good way? The positive effects identified in both research and healthcare are easily lost if there is no structure in place for long-term evaluation, follow-up and dissemination.

Patient and next-of-kin involvement is too often treated as a necessary evil or a box to be casually ticked, rather than a central, value-creating activity. Such an approach leads to initiatives that are neither particularly sustainable nor produce clear results, and a vicious circle is created.

There is a need for cultural and structural change regarding patient and next-of-kin involvement, both in research and in the healthcare sector. Structural change means that organizations need to review leadership, administration, financial systems and the way in which operations are structured. Culture change largely concerns the mindset and attitudes of individuals. Structure and culture need to work together and complement each other to maximize the benefits of collaboration.

Everyone who wants to collaborate must do their part. A good starting point is to read this report, which builds on the extensive research and rich experience already available in the field of collaboration.

If you are short on time

Pages 21–24 contain a compilation of recommendations for successful collaboration between stakeholders with different conditions, resources, knowledge, needs and objectives. The recommendations apply to research and innovation in the areas of research, healthcare and social care. They are summarized on one page in **Appendix 1**, with an associated checklist in **Appendix 2.**

But keep in mind that...

...there is no one-size-fits-all solution to collaboration. You must always adapt your tools to the context. You might even want to try designing your own toolkit or framework. In addition to those noted above, there are some additional appendices that may be helpful. **Appendix 6** includes several systematic and scoping reviews that may be relevant if you'd like to delve deeper. You can also take a closer look at **Appendix 7**, which contains an overview of some frameworks and toolkits suggested by different types of collaborative studies.

Preface

An international culture change is underway in collaboration, not least in the life sciences. It is clear that an inclusive approach is being increasingly prioritized, both in healthcare and in medical and care science research. Study participants, patients and next of kin are regarded as partners in an increasing number of contexts.[1–24] But what can we do to ensure that such partnerships are successful? And what exactly is meant by success in collaboration? These are the questions we try to answer in this report.

In 2020, a dialogue was initiated between ATMP 2030, Biobank Sweden, Genomic Medicine Sweden (GMS) and patient and next-of-kin representatives from the Swedish Network against Cancer, Rare Diseases Sweden, the Swedish Brain Tumor Association and the Swedish Lead Patient Network. All saw a need for more collaboration in the life sciences, and an informal working group was established.

The working group devoted 2020 to unbiased discussions on needs and wishes. At the end of the year, Biobank Sweden's Steering Committee decided to fund a one-year pilot study focusing on developing recommendations for sustainable and democratic collaboration with patients and next of kin. The pandemic caused delays, and ATMP 2030 and GMS financed an extension of the project during the first half of 2022, ensuring that this report could be finalized. A reference group comprised of 20 people representing healthcare, research, stakeholder organizations and public authorities in the life science field reviewed the first version of the report. **Appendix 3** contains a summary of those issues the reference group believed would be valuable to discuss and research further, but that are beyond the scope of this report.

One of our first steps during the pilot study was to define a common vision for collaboration.

OUR VISION IS:
a society more sustainable in the long term, where all members are able to contribute and to achieve the best possible health and well-being over time.

The report contains recommendations to stimulate and support this vision. One important aim of this is to contribute to crucial cultural and structural changes. The recommendations are based on proven experience and science. We hope that they can lay the foundation for successful collaboration between stakeholders with different conditions, resources, knowledge, needs and objectives, to enable research and innovation in the fields of medical and care science research, healthcare, and social care.

We also want to emphasize that the report itself is only one step in a journey of change. It adds the most value if it is used as a basis for further discussion and action, preferably together with the comments and questions in **Appendix 3**. We would like to especially thank the reference group for their time and commitment.

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Structure and limitations

The report is based on:

- an extensive review of scientific literature and reports
- so-called "gray literature," i.e., popular science literature or other literature that has not been subject to peer review
- a series of interviews with patient and next-of-kin representatives
- intra-organizational case studies
- several years of dialogue focusing on needs and challenges

Condensing such a large and diverse body of information into a format that is easy to communicate is a challenge, which is why we have gathered the key recommendations in the **Recommendations for collaboration section**. They are summarized on one page in **Appendix 1** and supplemented by a checklist in **Appendix 2.**

Another major challenge is that of limitations. There are many different forms of collaboration. Together, internationally recognized concepts such as patient and public engagement, co-production, community-based participatory research and the like constitute a large and complex field of research. Sometimes the corresponding terminology in other languages is used inconsistently and sometimes it is missing altogether.

The **Methods** appendix describes the material in greater detail, as well as the selection and working methods. In the **Definitions and concepts** appendix, this is supplemented by a compilation of common terms used in the area of collaboration. Quotes from the interview series are woven throughout the report. There are several systematic and scoping reviews that may be relevant for those who wish to delve deeper. A selection can be found in the appendix **Summary of studies**. Another compilation in appendix form is **Frameworks and toolkits**, which includes an overview of some frameworks and toolkits generated by collaborative studies.

The report's target audience is broad, with an emphasis on

- professionals, funders and decision-makers in healthcare, social care and research
- the general public, patients, next of kin, and the organizations that bring these groups together

Industry or other private actors have not been deliberately excluded, but as the working group does not have clear representation from, or experience of engagement with this part of the system, the focus of the report has been adjusted accordingly.

Collaboration between patient and next-of-kin representatives and industry brings with it specific challenges. Guidance for this sort of collaboration has been developed, for example, by the European Patients' Academy on Therapeutic Innovation (EUPATI).[25]

The authors of this report are involved in translational research. This means that results from experimental research are transferred to healthcare to benefit patients, or that observations or problems in healthcare give rise to new research ideas. In translational research, it is common to adopt a broader perspective on the concept of collaboration, which spans many contexts.[26] Hence, no distinction is made in the report between collaboration in medical and care science research and collaboration in healthcare. Many of the reviewed sources focus on more general collaboration perspectives. A number of others argue that it is counterproductive to distinguish at all between different collaboration concepts, as this makes it easy to lose focus on the actual goal: value-creating collaboration for all stakeholders.[26, 28]

In medical and care science research and in healthcare and social care, the general public as well as patients and their next of kin are common partners. These are three different groups with different conditions and needs, all of which possess expertise that is valuable in this context.[17] All of these perspectives are included throughout the report. There is no in-depth discussion of the patient as a consumer rather than a user, which is a distinction made in some theoretical models.[27]

Collaboration with patients and next of kin can take place at many levels, ranging from the individual to the group and society levels. The scope ranges from the individual care encounter to cross-organizational steering groups, from individual research studies to European or global research programs. In this report, we have chosen not to focus on any particular level. The specific challenges may differ depending on the level at which collaboration takes place. The recommendations in this report can be used at any level but should be adapted to the context.

Despite the report's focus on medical and care science research, healthcare and social care, the content can be applied to many types of collaboration between several different types of stakeholders with differing conditions.

Opportunities and challenges of collaboration

Engagement with the general public, patients and next of kin in medical and care science research has been highlighted as something that can provide meaningful outcomes that are useful to clinicians, patients and their families. In addition, collaboration can help build trust in relationships between patients, next of kin and healthcare professionals, particularly if trust has previously been poor or damaged.[8, 29, 30]

Efficiency, quality and patient safety are often emphasized as positive effects of collaboration in the healthcare sector. Collaboration can also help provide the individual with a better overall picture of the entire care process.[31–35] In translational research, collaborative initiatives are also highlighted as a particularly important success factor.[36]

The literature identifies several barriers to effective collaboration. Two main areas are highlighted as particularly problematic in terms of implementing successful collaborative initiatives:

- the lack of infrastructure that enables collaboration
- barriers to building and maintaining relationships within the collaborative initiative

The problem with relationships may be due, for example, to preconceived ideas about collaboration or a lack of trust between the stakeholders.[10]

The literature emphasizes that both a culture and a structure for collaboration need to be in place for it to work optimally. Below, we describe twelve aspects of collaboration, with examples of what works, what benefits it brings, what does not work and what obstacles exist. These aspects of collaboration are summarized in the recommendations. We have assumed that the same basic principles for successful collaboration apply in research, healthcare and social care.

Collaboration to define more relevant issues and objectives

There are risks in relying too much on (for example) organizational or budgetary needs and objectives in research and healthcare. Narrow perspectives lead to limited opportunities for value creation and innovation. One study shows that organizations

with representation from many different perspectives and an inclusive leadership find ten times more solutions to problems than organizations with less diversity.[37] Inviting and opening up for co-creation can be a way to engage marginalized groups and minorities that may otherwise be challenging to include in scientific studies, for instance.[38–40]

Establishing a clear structure and defining clear objectives facilitate the development of relevant issues to be addressed. Just casually including the patient and next-of-kin perspectives without reflecting on the shared benefits can have negative effects.[41] Moreover, the aims, objectives and form of the collaboration need to be clearly defined – in dialogue with everyone who is intended to participate.

Outcomes and effects of collaboration

It is clear from the literature that an important question to answer is: What outcomes and effects does patient and next-of-kin collaboration lead to? In many healthcare and research contexts, the value of such collaboration has been questioned. The question has been raised, for example, as to whether there are data to support that collaboration has an effect. A necessary follow-up question, then, is which data should answer this question and what should be used for comparison?

The effects of collaboration are often reported in terms of how the parties involved have experienced the collaboration initiative. When the literature discusses follow-ups to, and results of, collaboration, it is usually precisely those factors that are difficult to measure quantitatively that are emphasized. This may, for example, involve strengthened relationships and improved communication between the collaborating groups. But there are many types of outcomes that can actually be measured, such as adherence to treatment and better understanding of health-related factors, both of which lead to better health economics.[43–45]

In addition, at all levels of medical research, relevance to patients is a particularly important aspect.[46] It has been reported that collaboration leads to improvements in the design of study protocols or the development of relevant outcome measures.[47] In the case of drug development, it is also stressed that it helps if a sustainable framework for collaboration is in place from the outset when developing a targeted profile for the product, or recruiting study participants. [23, 48]

> *Those of us who are patients or next of kin with a high level of engagement move throughout the healthcare system in a way that the professionals do not. We not only see our own doctors and other healthcare professionals, but also sit on patient councils with regional and healthcare organizations and authorities, and in meetings with the Government and Parliament. We are therefore able to see more comprehensive solutions. For example, I have been able to identify unnecessary care on several occasions.*
>
> Quote from interviews with patient and next-of-kin representatives

From symbolic collaboration to co-creation

In order to achieve the desired effect, those who wish to collaborate must reflect on the level of collaboration that needs to be carried out.

> Collaboration with patients or next of kin is sometimes criticized for being symbolic rather than genuine. This may be because it is often implemented at a purely **consultative** level, i.e., simply collecting or disseminating information to patient and next-of-kin representatives, without involving them further in any decision-making process or follow-up.
>
> The next level, the **cooperation** level, may involve inviting the representatives to the actual implementation stage of an intervention, but without prior involvement in the design of the intervention in question.
>
> At the **co-creation** level, patient and next-of-kin representatives are actively involved throughout the intervention.[43, 49–51]

A common form of collaboration in many countries consists of various types of patient or next-of-kin councils. Councils as a method can very well contribute to cooperation and co-creation, but unfortunately often stops at purely consultative or symbolic collaboration.[52]

Attitudes towards collaboration and its costs

Clinicians and researchers may find collaborating with patients and families challenging, sometimes even as something that negatively impacts their work and requires additional costs or resources.[28, 53–55]

In a 2018 UK study, researchers highlighted several obstacles to collaboration, including:

- lack of financial support
- lack of support from leadership within the organization
- lack of administrative support
- emotional labor and emotional stress in general
- insufficient room for collaboration within the prevailing "publish or perish" culture in academia[54]

Despite the obstacles, however, even the critical voices state that they believe in the value of collaboration if it is carried out methodically and with a well-defined purpose. It is also reasonable to assume that preconceived notions and outright myths to some extent underlie the resistance to collaboration that has arisen at different levels and in different contexts.

The concept of shared decision-making in healthcare is an example of just such a collaborative model that has met with resistance. Arguments against shared decision-making include:

- that it would take too much time
- that the patients themselves are not interested in participating
- that it is not an appropriate model for the majority of decisions made in patient care
- that this type of collaboration would be impossible to implement due to quality concerns and similar reasons

However, evaluations show that this criticism does not reflect reality. Within the framework of current guidelines for making decisions about individual patient care, time is already set aside for shared decision-making. There is no evidence to suggest that a majority of patients would be disinterested in this. Above all, evaluations show that shared decision-making has consistently improved clinical outcomes and further stimulated engagement among patients.[56]

Trust, time and sustainability

In order to build genuine collaboration, a partnership is needed, which requires trust – and trust is something that needs to develop over a long period of time.[57, 58] Therefore, a long-term approach is a key success factor. This is also something that patient and next-of-kin representatives themselves highlight as particularly valuable in the dialogue with researchers.[59]

However, representatives in the working group behind this report have noticed something interesting in dialogue with various councils for patients and next-of-kin and the

general public. Continuous collaboration in small groups can lead to an excessive striving for consensus or loyalty that can counteract the purpose of collaboration. This is a potential obstacle that deserves special attention. A long-term approach and continuity therefore need to be supplemented with continuous evaluation of the purpose of the collaboration and who/which organization is being represented. This applies to patient and next-of-kin representatives as well as to professional representatives.

It is common for collaborative initiatives to be temporary efforts, such as projects, rather than measures that are permanently realized in existing structures and systems. This means that any influence is also only temporary.[1, 60] Structural changes are needed in the short term and can lead to culture change in the longer term.

Transparent communication is also important in terms of building trust and is an essential component of any collaborative initiative.[61–64]

> *Building good collaboration together requires more power and energy than most people are willing to put in.*
>
> Quote from interviews with patient and next-of-kin representatives

Power (im)balance and valuing of expertise

There must also be a clearly defined justification for what collaboration is needed, why, and between whom.[29] The selection of the parties contributing to the collaborative initiative must therefore be based on aims and objectives. If the aims, roles and expectations are clearly defined, the risk of creating a power imbalance is reduced.

The criticism of collaboration as something that only has a symbolic function is often an effect of a power imbalance in the collaboration structure. For example, this may involve only certain parties having decision-making authority, or certain stakeholders being completely excluded.[43, 49, 65, 66] In order for collaboration to gain a foothold in an organization, such as a hospital, management must take an actively supportive

and motivating stance. In addition, the teams working on collaborative projects must continuously develop the initiatives and their tools.[12] This can also be called a balanced top-down, bottom-up approach. If only the top-down management perspective prevails, this will not lead to good collaboration results.[67]

It can also be the case that information only flows in one direction. An example of this is if it is only the patient, next-of-kin representative or other member of society who is to be educated or receive information. A flow that goes in the other direction is absent in a surprisingly large number of cases.[9, 29, 68] This creates a power imbalance in terms of which party possesses the most highly valued knowledge, where the expertise that is most highly valued is usually that of the professionals.[29, 30, 69–71] Many people forget that expertise can take many different forms. Patients' expertise can be of different types that are relevant to different parts of a collaborative process, for example in research.[42] In addition to their knowledge and experience of their own or their family member's illness, patients and next of kin also bring experiences, skills and knowledge from working life and education that should be surveyed and utilized. One successful measure has been to openly recognize and acknowledge the issue of power imbalances, thereby integrating discussions on power dynamics into the collaborative form.[30] A further step is power sharing, i.e., taking concrete measures to reduce or completely eliminate power imbalances.[39] A comprehensive study focusing on collaborative initiatives has shown that patients, next of kin and other representatives of society are usually excluded if they are not clearly defined as stakeholders from the outset.[65]

> *One of the best examples of collaboration I have been involved in was at the management table of one of the larger university hospitals. Here, the professionals really wanted to understand what the challenges were and the views of us patient and next-of-kin representatives were taken on board, which led to change. A key factor was building trust between everyone at the table by addressing common challenges and valuing everyone's experience equally.*
>
> Quote from interviews with patient and next-of-kin representatives

At the same time, studies show that healthcare and research initiatives entirely led by patients have been criticized for creating an imbalance in the other direction.[72] Reciprocity in collaboration is thus of great importance so that researchers and clinicians do not feel excluded or reduced to the role of bystanders. The lived experience does not replace other perspectives and competencies in a research or

healthcare team – it complements and expands them.[73] For example, in cases where patient councils are established, they have significantly better outcomes if there is solid representation from the clinical side as well.[74] The Patient-Centered Outcomes Research Institute (PCORI) is an example of an organization that has implemented a process to include both researcher and patient (or other stakeholder) perspectives in a sustainable way.[75]

There is often a slippage in representativity or perspective when lay people are transformed into patient or next-of-kin *experts*. This can happen because they gain a greater degree of knowledge about, for example, the healthcare system or the research system, which means that the boundaries between the different identities can become unclear. Those who are expected to represent the patient's, next of kin's or another's perspective may instead adopt a system perspective, such as that of the healthcare, social care or research system.[29, 76]

> *One of my worst examples is my time as a patient representative in the development of a healthcare organization. Everyone was so terribly uncomfortable that there was a patient at the table. Finally, I put my foot down. Slowly, slowly there was a change, but it took a year before I felt at ease, and as part of the group. It was clear that there was a hierarchical structure and that a culture change was needed.*
>
> Quote from interviews with patient and next-of-kin representatives

Representativity in collaboration

A further power imbalance arises if representativity fails, leaving some groups underrepresented.[43, 67, 77] This is particularly problematic in a healthcare context, where some groups are simply unable to represent themselves.[14, 17, 78, 79] Another representativity challenge relates to accessibility. Patient and next-of-kin representatives who choose to participate in collaborative initiatives often have a high level of commitment. This is a positive characteristic that is important for successful collaboration. However, it can also mean that value-creating perspectives are completely excluded.[1, 80] Patients who are particularly frail or seriously ill are a key group. Studies show that their contributions include improved study design and more relevant research, but that there are particular challenges to collaboration, as they may be less accessible and flexible than others.[14] It is therefore important to assess specific risk factors related to representativity from the outset and be prepared to take steps to address any issues

that arise. In studies involving marginalized groups or minorities, communication can be a crucial factor. Targeted and thoughtful communication can significantly influence study outcomes.[81] In groups with weaker communication skills, adjustments such as commonly agreed channels and modes of communication may even improve overall communication skills.[17]

The working group believes that a distinction should also be made between representativity in terms of personal perceived experience and democratically assigned representativity. Context and purpose determine which type of representativity is appropriate.

Remuneration for participation and work

A valued resource should of course receive compensation. This is something that patient and next-of-kin representatives see as a necessity to ensure commitment in both the short and long term.[73, 82, 83] However, it is not uncommon for patient or next-of-kin representatives to be expected to participate without any form of remuneration for their efforts. This is often related to the lack of definition of overall aims, roles, or what the representatives are expected to contribute. The general rule is that all work done by patients and next of kin should be fairly compensated, in the same way as for other stakeholders. However, flexibility and dialogue may be required regarding the appropriate form of remuneration for participation, especially if the group of patients or next of kin you want to interact with is living on income support or sickness benefit. In such situations, hourly remuneration or other forms of financial compensation may have a negative impact on the participant's livelihood and be inappropriate, which is why other compensation options need to be considered.[1, 69, 83] For example, it is not uncommon for patient and next-of-kin representatives to be offered gift vouchers as an alternative to financial compensation.

Increased interest and impact

One important effect of collaboration is that public interest in research increases, which can in turn lead to wider dissemination of research results, more external support, greater impact, and better research quality.[60, 66, 84] But this requires working on quality issues. The better the framework for collaboration that is in place, the more relevant the research questions will be to the public.[85–87] Patients have expressed that they participate in order to give back to society.[82] They also believe that they have influence over societal developments, which increases perceived justice, system confidence and trust in the democratic process.[21, 77] However, it is important to emphasize that the question of why patients, next of kin and other representatives of the public engage in collaboration in the healthcare area, is still relatively unexplored.[88, 89]

Increased opportunity for funding

The conditions for cooperation vary from country to country. The United Kingdom, the United States, Canada and Australia are particularly advanced in terms of regulating collaboration with patients and next of kin.[75, 90–92] In recent years, the UK has also made increasing demands for collaboration with patients and next of kin in various research contexts on the grounds that it makes research more effective, more credible and often more cost-effective. Even now, collaboration is a necessary element for research projects to be considered at all by some research funders, and a strategic national goal in the UK is to make collaboration a crucial part of all excellent research.[2, 31, 54] Measures have also been taken to ensure stability and a long-term approach. Similar initiatives also exist on a large scale in Canada, the US and Australia.[90] However, it is important to emphasize that research conducted with collaborative elements generally contributes to increased chances of obtaining funding even if there are no formal requirements for collaboration.[47]

Evaluation and follow-up

Much of the literature emphasizes that we lack follow-up on the outcomes of collaborative initiatives and that this is one of the most important measures that needs to be taken, particularly in order to identify which types of interventions are most effective.[29, 41] A common problem is that evaluations of patient and next-of-kin collaboration do not follow any established framework for assessment or follow-up.[29, 41, 43, 93] Not being able to evaluate or follow up on outcomes naturally contributes to the complete absence of greater benefit and pervasive impact. Here, however, it is important to emphasize that this does not mean that the results themselves are inadequate.[16] As discussed above, collaboration has been criticized for having only a symbolic function, for example in terms of representativity and diversity, but this is something that completely disregards all the documented positive effects of collaboration.[43, 49, 70, 94, 95] Collaboration being perceived as symbolic is often linked to the failure to define aims and objectives in cooperation with the parties involved. In the cases where follow-ups are performed, these are often insufficient. For example, the focus might be on how research study participants themselves are affected by participating in an initiative for patient and next-of-kin collaboration, rather than on how their involvement affected the study design or outcome of the research.[96]

An important aspect of evaluation and integration can be the production of policy documents and documentation.[97] However, it is important to design these so that they do not become a further obstacle on the path to good collaboration. A major study of existing guidelines for collaboration showed that they were often based on a normative and inadequate perspective, in which central problems such as power imbalances were not included at all.[98] Recommendations for how documentation should be designed are also available for research funders and ethics committees.

The documentation completed by researchers prior to a project should integrate and involve patients more broadly and over the entire life cycle of the study.[99]

The results of collaboration should be measured against clear evaluation criteria agreed on in advance by all parties. The same applies to the principles of follow-up and dissemination. It is not uncommon for potential findings to be forgotten, or in some cases completely swept under the rug.[29]

> *My experience of being on a patient council is that they talk and I listen. It often feels like a box to be ticked – the inclusion of a patient representative. An example is government assignments where authorities invite us in, but where there is little room to actually deal with the views of patient and next-of-kin representatives. I believe that part of this is due to the fact that the government assignments themselves are rigid, with little opportunity for the authorities to control the issue. Comments from me or others are rarely included in the proposals for change. Even if the meeting with the patient or next-of-kin representative leads to good ideas, there is still no opportunity for the authorities to do anything with these ideas. Collaboration leads to nothing more than dialogue, which is certainly inspiring, but has no effect. The council is then perceived as merely providing information to patient and next-of-kin representatives, with no ability to exert influence.*
>
> Quote from interviews with patient and next-of-kin representatives

Disseminating results, methods and frameworks

Studies show that flexible collaborative models that can be adapted to specific conditions and needs are more useful.[100, 101] It is unrealistic to assume that one framework or toolkit could be applied to all contexts. At the same time, it is important to emphasize that overlapping initiatives and constantly reinventing the wheel have an erosive effect on collaborative work.[102] The majority of the frameworks developed in medical research, for example – and there are many – have not been used outside the specific context in which they were developed.[100] This may certainly be due to the narrow context in some cases, but in the majority of collaborative models, it is probably more a question of a lack of knowledge of what work has already been done. This, in turn, may be due to a lack of goal-setting – if dissemination of experience is not

recognized as an important aspect of the culture change everyone needs to contribute to, it is not surprising that it is forgotten, or that a report or article is written in a way that makes it difficult for someone else to re-use the knowledge.

The lack of dissemination may also be linked to the problem of power imbalances. As one study points out, patient-driven innovations are rarely disseminated beyond a narrow, personal area of use.[3] This can be compared with the tens of thousands of scientific publications focusing on collaboration, based on contributions from a single group of collaborative partners lacking representativity. Here, too, there is a need to work together to purposefully define and design the dissemination of results and experience.

Summary

The positive effects of collaboration presented in the studies discussed above can be summarized as that collaboration:

- builds trust, both in collaborative environments and in the system (see p. 10)
- contributes to efficiency, quality and patient safety (see p. 10)
- provides a better overall picture of the entire care process (see p. 10)
- contributes more perspectives and thus more potential solutions to problems (see p. 10)
- leads to better healthcare economics (see p. 11)
- contributes to improved study protocols (see p. 11)
- provides more relevant outcome measures (see p. 11)
- leads to easier recruitment of, for example, study participants (see p. 11)
- creates greater involvement in patients' own care (see p. 13)
- contributes to increased public interest and support for research (see p. 17)
- leads to wider dissemination of research results (see p. 17)
- leads to better research quality (see p. 17)
- provides increased opportunities for research funding even in the absence of formal collaboration requirements (see p. 18)

How can we achieve well-functioning collaboration that creates added value? The following six recommendations aim to promote and support the vision of *a society more sustainable in the long term, where all members are able to contribute and to achieve the best possible health and well-being over time*. It is clear that the collaborative initiatives that have delivered good results and added value have identified and addressed the pitfalls and challenges discussed in this report. They have also worked with both culture and structure to create functioning, long-term collaboration. The following recommendations are building blocks for raising collaboration with patients and next of kin to the 2.0 level. The more building blocks that are used, the better the results that can be achieved.

Recommendations for collaboration

1 Define clear aims, objectives and evaluation criteria – together

Collaboration 2.0 focuses on co-creation rather than consultation, i.e., on co-creating *with* patients and next of kin rather than *for* them. The work should begin with the definition of shared aims and objectives and the establishment of a process for joint evaluation and follow-up. Policy documents can support the process, but their development should also be based on principles of co-creation.. Another thing to consider is that research, innovation and care processes are continuously changing and interconnected. The evaluation of one initiative often forms the basis for planning the next, and collaboration must be continuously integrated, evaluated and developed. The composition and role allocation of the group also need to be based on such principles – it may become clear at an early stage that the people or organizations represented in the group need to be adjusted, and sufficient flexibility is needed to address this. To maximize value creation, a balance is also required between a long-term approach and trust, and new approaches and a questioning of the collaborative work. This may mean that time limits need to be set for how long the collaborators will act as representatives.

Can help ensure that:

- Collaboration is evaluated and monitored in terms of its aims, objectives and effects
- The problem of symbolic collaboration is reduced thanks to a clear collaborative framework
- Partners with the right competencies for the task are included in relevant parts of the process
- Negative effects of perspective drift are reduced through clear roles

2 Confront power imbalances

Good collaboration builds on a foundation of complementary and well-calibrated competencies. The issue of power balance in terms of which perspectives are valued – and how they are valued – should be discussed at an early stage. There must be openness about how everyone in the group views each other's expertise. When this is addressed, it is also possible to contribute to increasing each other's knowledge and expertise by creating shared learning. Sometimes the starting points in a collaborative group can be so different that measures need to be taken to adjust the power imbalances. Even if the composition of the group is based on the aims and objectives of the collaboration, a preparatory training program or other form of knowledge-enhancement activity may be needed for some or all of the parties involved.

Can help ensure that:

- An equal and inclusive environment is created, where there is a balance of power rather than an imbalance of power
- Trust in the expertise of the other collaborative partners is enhanced
- All parties are actively involved and it is clear who is expected to contribute what
- Knowledge exchanges take place between all parties and everyone is given the opportunity to develop their skills

3 Communicate to build trust and confidence

Trust and confidence are key components of effective collaboration, and for these to be possible, clear and transparent communication is essential. Communication should be a two-way channel with an openness to take in all perspectives. There is also a need to reflect on the purpose of the communication. If views are collected in order to implement a change process, these views cannot simply be thrown on a pile without any changes being made. Regular communication is necessary if this is not already an integral aspect of the collaborative model. Questions need to be answered and both positive and negative views need to be discussed openly. It is also important that communication is adapted for all relevant target groups, which in many cases may have different needs.

Can help ensure that:

- Long-term relationships within the collaborative initiative are built and maintained thanks to trust-building communication
- All perspectives and views are gathered through an open flow of information in all directions
- A culture change takes place, thanks to the building of trust and confidence
- Challenging problems get new potential solutions through open dialogue

4 Create conditions for remuneration and representativity

Remuneration must be paid to all collaborating parties. A remuneration model must therefore be in place and agreed upon before any cooperation is initiated. The nature of remuneration and other support can also affect who is able to represent. For example, not everyone has the same opportunities to receive certain types of remuneration or to attend a physical meeting. In order to create the best possible representativity, remuneration and other forms of support need to be reviewed. The representatives who voluntarily sign up for a collaborative initiative do not always fulfil all the requirements for representativity, but they are active and engaged and important to bring in for that reason. As a complement, and to reach minorities and marginalized groups, for example, outreach activities may be necessary, even if they are more resource- and time-consuming. This applies to patient and next-of-kin representatives as well as to professionals. Another important aspect to consider is scheduling meetings and other joint work to suit all participants in the collaborative initiative, including, for example, frail and seriously ill individuals. It cannot be assumed that everyone has the time, desire or energy to act as a full-time representative, or that everyone has access to the same tools for successful communication.

Can help ensure that:

- Power imbalances are reduced or eliminated
- More perspectives can be included through the introduction of flexible remuneration models
- Key stakeholders who would otherwise be missed can be included through adaptation to individual needs for participation
- Trust and belonging are promoted through the introduction of an inclusive way of working

5 Build a long-term structure for collaboration

Short-termism is highlighted as one of the main obstacles to successful collaboration. In order for a culture change to take place in collaboration, it is therefore necessary to aim beyond individual projects. This can be perceived as challenging when resources are limited and when timeframes and deliveries are determined based on the perspectives and needs of several other stakeholders. It often becomes a question of whether the benefits of collaboration outweigh the risks and costs. It is therefore important to work consistently for a long-term approach, and a prerequisite for this is that a structure for collaboration exists within the organizations and systems in which it is

intended to be implemented. Without a structure, for example in the form of processes, IT systems, allocated working hours, budgets, management systems and adapted leadership, collaboration becomes burdensome both administratively and in terms of time. A structure for collaboration is therefore necessary in order to develop a collaborative culture that is sustainable in the long term. The structure needs to be flexible enough to support collaboration based on defined aims and objectives.

Can help ensure that:

- Costs and time are reduced by not having to reinvent the wheel every time you need to collaborate
- The problem of purely symbolic collaboration is eliminated when collaboration is a permanent feature in the organization or system
- Recruiting representatives to different collaborative initiatives is easier because a structure is already in place
- Trust and confidence in an organization grow if the organization signals that collaboration is such a priority that a solid structure is in place
- Loss of knowledge and skills is reduced because there is a structure to manage them over time

6 Share positive and negative experiences and learn from others

An important part of collaboration is that all parties must take responsibility for sharing experiences, results and effects – both negative and positive. It often comes naturally to talk about what works well, but a central part of sharing is also preventing others from repeating mistakes already made. It is therefore important to create the conditions for continuous learning and for lessons to be passed on beyond the collaborative partners. If the benefits of collaboration are to outweigh the costs, it is essential to use what others have already developed and tested. You can then adapt successful methods and channels to your own activities. You should also reflect on how to communicate your results in the way that adds the most value.

Can help ensure that:

- Others are helped to collaborate better, leading to an improved culture of collaboration throughout society
- New cross-organizational and cross-system collaborations are made possible thanks to the wide dissemination of results and experiences
- A collaborative approach is normalized through open dialogue
- Public understanding and interest in collaboration increase

Beyond the recommendations

There is much you can do yourself in terms of building both culture and structure to support your collaboration. But there are important drivers beyond the recommendations above. Some areas we particularly want to highlight to enable the next stage of collaboration with patients, next of kin and the general public are funding, legislation, and collaboration in product innovation.

Research funders

It is not necessarily the case that patients and researchers have such differing views on what should be prioritized in research, or even on what patient and next-of-kin involvement should look like. A British study from 2012 found that researchers and the public were in nearly complete agreement about the areas that were possible for collaboration.[103] At the same time, collaboration means that researchers potentially need to reflect on their approaches and perhaps need to re-evaluate their views on such things as expertise and knowledge.[69] Studies have shown that researchers tend to underestimate the relevance of contributions from collaborative representatives, as well as the extent to which they themselves use these contributions in research planning.[104] It is thus a question of the need for a culture change, rather than targeted, isolated measures. For such a culture change to be feasible, it is necessary that everyone feels it is both meaningful and necessary.[90, 105, 106]

A common problem is that temporary collaborative solutions are only implemented to solve a problem, rather than being integrated as a permanent part of the system.[43, 107] Dialogue with research funders is a particularly important piece of the puzzle in this regard. Studies show that research funding bodies have an important and influential role in terms of how collaboration is defined and interpreted among health and medical researchers.[4] Some argue that the implementation of best practices will not have an impact until research funders are committed to building and maintaining an effective collaborative structure.[20] This includes, for example, integrating compensation for such collaborative representatives into funding models.

It is clear that many countries have come a long way in terms of systemic change and integration of patient and next-of-kin perspectives in current research funding structures. Those who have not, should take measures to not be left behind.

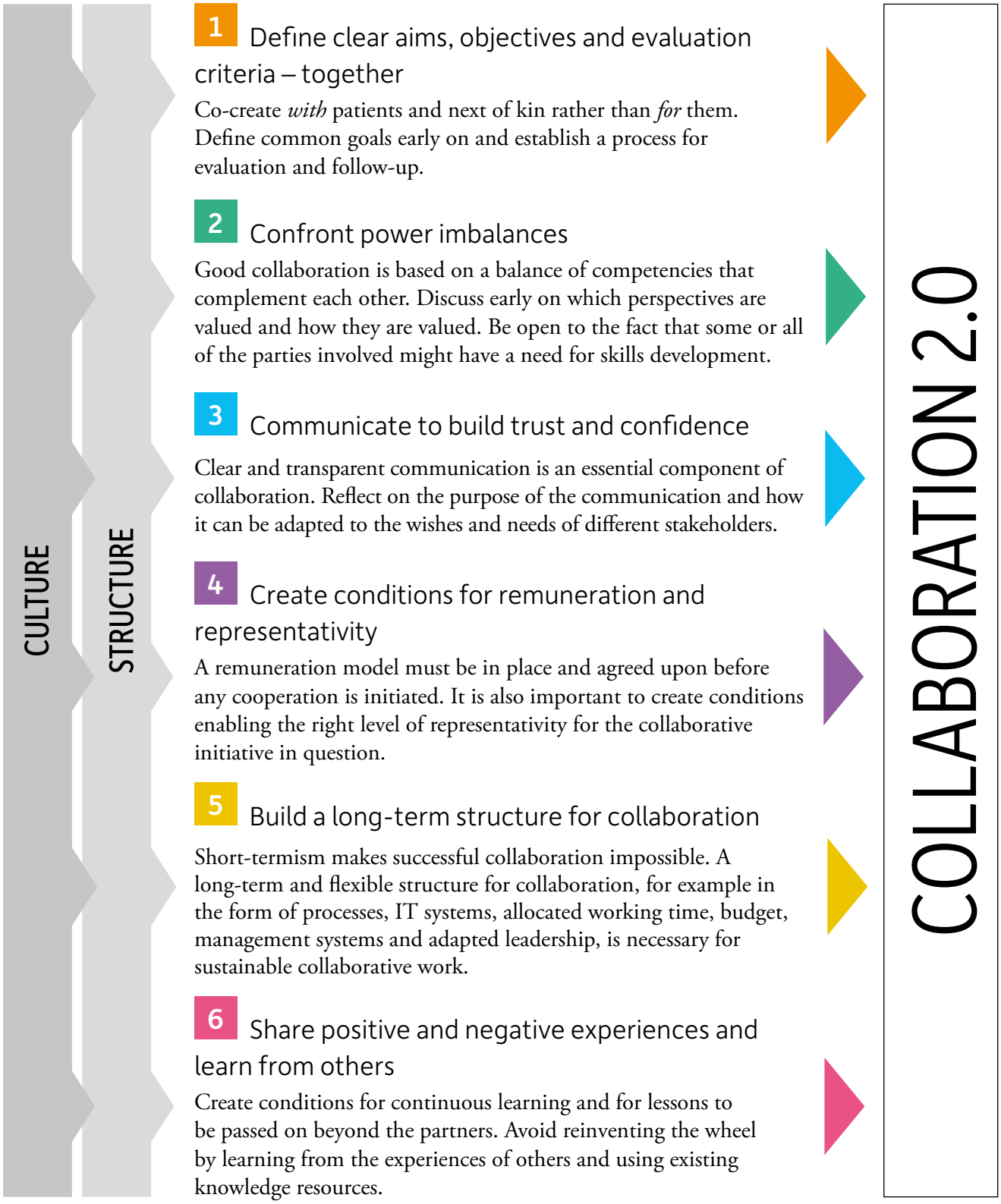
Legislation on remuneration structures

Flexible, long-term structures and opportunities for remuneration for active participation are some of the key components of effective engagement with patients and next of kin highlighted in the report. Democratically assigned representativity, i.e., acting for a patient or family member group as a collective, is an important role for patient and/or next-of-kin associations and also something that is needed in many contexts. One question that must be addressed in relation to this is how patient and/or next-of-kin associations receive state support for their work. In the Swedish context, the Agency for Health and Care Services Analysis's report (2015:4) *Sjukt engagerad* [Sick engagement] mapped the patient and disability movement.[108] It highlighted how the conditions for patients' collective participation in the design of healthcare could be strengthened. In 2016, the Swedish National Board of Health and Welfare was commissioned by the Swedish Government to review Ordinance (2000:7) on support for disability organizations.[109, 110] In December 2017, the National Board of Health and Welfare submitted a legislative proposal with comments to the Government (Ref. S2016/07041/FST). This proposal describes how the National Board of Health and Welfare collaborated with the disability movement, including through surveys, forums and presentations in the so-called "big council" (a council of organizations that have been awarded government grants for disability organizations for the grant year in question). Many smaller organizations asserted positions during the course of the assignment, but have not received any feedback on the outcome of the assignment. The legislative proposal has not yet led to any amendment to the ordinance. In brief, Ordinance (2000:7) on support for disability organizations currently limits the possibilities for government grants to so-called "disability organizations" that have more than 500 members and a nationwide presence with local or regional roots in at least ten counties. This limitation makes it difficult for organizations that, for example, have many members (over 500) but do not have ten local associations, or the opposite – fewer members but many local associations. An updated and more flexible remuneration structure in terms of both the number of members and local associations is requested by, among others, the patient and/or next-of-kin representatives who participated in the preparation of this report, as they feel that the current ordinance leads to inequalities in state support.

Collaboration in product innovation

As described above in terms of the structure and limitations of the report, frameworks for collaboration with companies in research or innovation processes have not been specifically addressed here. However, we recognize that product innovation in collaboration with industry is an important part of research and development in both healthcare and social care. As previously mentioned in the report, EUPATI has developed a toolkit with guidance and training activities.[25, 111] Work is carried out on the national level as well, such as the version developed by EUPATI Sweden.[112] An important step in the future development of Collaboration 2.0 will be to investigate how the content of this report relates to this and other published material on patient and next-of-kin collaboration in product innovation.

Appendix 1: Overview of recommendations



Appendix 2: Checklist of recommendations

Recommendations	Already doing	Will review
1 Define clear aims, objectives and evaluation criteria – together **Can help ensure that:** • Collaboration is evaluated and monitored in terms of its aims, objectives and effects • The problem of symbolic collaboration is reduced thanks to a clear collaborative framework • Partners with the right competencies for the task are included in relevant parts of the process • Negative effects of perspective drift are reduced through clear roles	☐	☐
2 Confront power imbalances **Can help ensure that:** • An equal and inclusive environment is created, where there is a balance of power rather than an imbalance of power • Trust in the expertise of the other collaborative partners is enhanced • All parties are actively involved and it is clear who is expected to contribute what • Knowledge exchanges take place between all parties and everyone is given the opportunity to develop their skills	☐	☐
3 Communicate to build trust and confidence **Can help ensure that:** • Long-term relationships within the collaborative initiative are built and maintained thanks to trust-building communication • All perspectives and views are gathered through an open flow of information in all directions • A culture change takes place, thanks to the building of trust and confidence • Challenging problems get new potential solutions through open dialogue	☐	☐

Recommendations	Already doing	Will review
4 Create conditions for remuneration and representativity **Can help ensure that:** • Power imbalances are reduced or eliminated • More perspectives can be included through the introduction of flexible remuneration models • Key stakeholders who would otherwise be missed can be included through adaptation to individual needs of participation • Trust and belonging are promoted through the introduction of an inclusive way of working	☐	☐
5 Build a long-term structure for collaboration **Can help ensure that:** • Costs and time are reduced by not having to reinvent the wheel every time you need to collaborate • The problem of purely symbolic collaboration is eliminated when collaboration is a permanent feature in the organization or system • Recruiting representatives for different collaborative initiatives is easier because a structure is already in place • Trust and confidence in an organization grow if the organization signals that collaboration is such a priority that a solid structure is in place • Loss of knowledge and skills is reduced because there is a structure to manage them over time	☐	☐
6 Share positive and negative experiences and learn from others **Can help ensure that:** • Others are helped to collaborate better, leading to an improved culture of collaboration throughout society • New cross-organizational and cross-system collaborations are made possible thanks to the wide dissemination of results and experiences • A collaborative approach is normalized through open dialogue • Public understanding and interest in collaboration increase	☐	☐

Appendix 3: Reference group and discussion questions

Reference group

Catharina Barkman (Forum for Health Policy), Britta Berglund (Rare Diseases Sweden), Anna Blommengren (Karolinska University Hospital), Mats Brommels (Karolinska Institutet) Helena Conning (Swedish Network against Cancer), Kristina Gustafsson Bonnier (Rare Diseases Sweden), Sonja Eaker (Biobank Sweden), Roger Henriksson (Umeå University), Eva Jolly (Karolinska Comprehensive Cancer Center), Siri Kautsky (RCC (Research Cancer Centres) Stockholm Gotland), Dag Larsson (Lif (Trade association for the research-based pharmaceutical industry)), Karin Lilja (Swelife), Frida Lundmark (Lif (Trade association for the research-based pharmaceutical industry)), Lisbeth Löpare Johansson (Swedish Association of Local Authorities and Regions), Anna Martling (Karolinska Institutet), Karin Mellström (Swedish Childhood Cancer Fund Stockholm Gotland), Anna Nilsson Vindefjärd (Research!Sweden), Peter Nordström (Swelife), Lars Palmqvist (University of Gothenburg), John Stewart (Swedish Network against Cancer).

After reading the first version of the text, the reference group highlighted several aspects that are important to consider and discuss in order to achieve successful collaboration, but that are beyond the scope of this report. These comments and aspects are summarized below and are intended to form a basis for further discussion.

– Joint responsibility too often becomes nobody's responsibility. How should we address responsibilities, powers, rights and obligations, for example in relation to the recommendations?

– Who is responsible for carrying out good and objective follow-ups?

– Do we want to collaborate with an organization in which one person represents many people or do we want to collaborate with individual patients? Are both needed?

– Who is responsible for clarifying who should represent what and why – and in what contexts?

– Patient and next-of-kin representation is often missing when high-level cross-boundary collaborations (industry, universities, healthcare regions) are established and evaluated. How can such a structure be put in place?

– Platforms that ensure knowledge exchanges between, e.g., different competencies and research areas are lacking in many cases. These would be good environments for collaboration with patients, next of kin and other stakeholders who are often otherwise excluded. How do we create such platforms?

– How do we deal with the complex levels of healthcare? At what levels should collaboration be prioritized and why?

– What are the effects of patient participation in the longer-term health economic perspective?

– How are specific types of collaboration or specific effects of collaboration linked to different overall socioeconomic benefits?

– How do we perform detailed evaluations of the impact of different types of collaborative initiatives on health and social economics?

– How do we best recognize and make visible the needs and views of groups who cannot represent themselves, such as children or people with severe cognitive difficulties, multiple disabilities and the like?

– How do we take a balanced approach to address the issue of confidentiality in different collaborative contexts where it may be a problem?

Appendix 4: Methods

Background and methods

In its earliest version, the project began in December 2019. At that time, Biobank Sweden asked other organizations in the life science field whether they were interested in working together on patient and next-of-kin collaboration. A small group of representatives from ATMP 2030, Biobank Sweden and GMS (Genomic Medicine Sweden) then began work on developing a structure for collaboration. Initially, the aim was to establish a consultative patient council or network. Four patient and next-of-kin representatives who were already connected to the organizations were invited to join the project, and the challenges and benefits of this type of collaboration were then discussed together, mainly based on the patient and next-of-kin representatives' experiences of being part of patient councils. It quickly became clear that a traditional, consultative patient council would hardly provide the desired outcome. In 2021, a one-year pilot study was therefore carried out in the framework of Biobank Sweden's research infrastructure, funded by the Swedish Research Council. The pilot study was called Collaboration 2.0 and the working group examined, through continuous meetings, workshops and individual work, different frameworks for patient and next-of-kin collaboration and what kinds of outcomes different approaches have resulted in. The pilot study was extended during the first half of 2022 with funding from ATMP 2030 and GMS, with the aim of producing a final report. The content of the report is based on a literature review of scientific literature and gray literature (popular science literature or other literature not reviewed by subject matter experts) and a series of interviews with patient and next-of-kin representatives.

Literature review

The working group decided to conduct a general literature review with systematic literature searches and agreed on two main questions to address. The term "citizens" in this context includes patients, next of kin, and all other representatives of society, independent of their citizenship status.

1. Citizen involvement in **health promotion and healthcare** – what works well and what to avoid?
2. Citizen involvement in **research** – what works well and what to avoid?

A central part of the work was to set up the search strategy to answer these two questions. With regard to the gray literature, no specific search method was used. The members of the working group suggested relevant reports and the like, which were

included or excluded after discussion within the group. For scientific literature, i.e., literature that has been peer-reviewed, systematic literature searches were used.

Here, the diversity of the working group, with patient, next-of-kin, research and healthcare perspectives, had a major impact on the results. All members of the working group jointly put together and read a selection of key articles. Based on the articles, English medical subject heading (MeSH) terms were identified and used as the basis for an initial search. All searches were carried out with the assistance of Sabina Gillsund, librarian at Karolinska Institutet University Library. Some fine-tuning was done after the first search. For example, the MeSH term *patient participation* yielded too many search results, as too many hits focusing on shared decision-making were included. As positive and negative outcomes were particularly relevant, the requirement was added that the terms *framework* or *model* must be included in the title or summary of the articles. The selection of search results was limited in time to articles published between 2003 and 2021. After the adjustments, the search was conducted in four scientific databases: Medline (Ovid), Web of Science Core Collection, PsycInfo (Ovid) and Cinahl (Ebsco). The full search strings can be found here: https://genomicmedicine.se/samverkan-2-0-litteraturstudie/.

The search string for citizen involvement in health promotion and healthcare initially yielded 5033 articles and after screening for duplicates, 3683 articles remained. The search string for citizen involvement in research initially yielded 5226 articles and after screening of duplicates, 4094 articles remained.

The total of 7777 articles were transferred to Rayyan (rayyan.ai). At least two members of the working group read each title and summary and marked whether the article should be included or excluded. The aim was to find an illustrative selection of examples on which to build the report, rather than to do a full analysis for a scientific publication. The final sample was therefore narrowed down to represent the clearest trends in the literature. 57 articles focusing on citizen involvement in health promotion and healthcare and 71 articles focusing on citizen involvement in research were included in the initial reference list. The sample was divided among the working group members and the data was extracted in tabular form and integrated into the report. The text was then processed during a series of workshops. After editing, 115 articles remain in the final version of the report. This also includes a few articles resulting from snowballing/bibliographic selection. Such articles were identified as particularly important by one member of the working group and were then proposed to, and where appropriate approved by, the other members of the group.

Appendix 5: Definitions and concepts

Patient and next-of-kin collaboration

One challenge is that there is no generally accepted terminology for the concepts used in the area of collaboration. It is possible in some cases to use MeSH terms, but many terms are missing. It can also be unclear how widely accepted certain terms are. There are also variations in the definitions of the terms. See the reference list for further details on areas of use and definitional problems.[28,30,43,54,56]

Patient and next-of-kin collaboration is the term used throughout this report to describe all forms of collaboration with patients, next of kin and other representatives of the public, regardless of area. It thus serves as an umbrella term for many related terms focusing on the interaction between many different actors in many different contexts. Below is a selection of such terms, their acronyms, and their definitions.

Term	Definition
Citizen science [113]	A type of research in which members of the public and researchers work together to develop new scientific knowledge.
Co-creation [84]	Indicates a type of research or healthcare program in which patients or other representatives of the public are involved in the design of the research and care.
Co-production[26]	Indicates a type of research or healthcare program in which patients or other representatives of the public are involved in the design of the research and care.
Community engagement (CE) [114]	Indicates an approach in which the healthcare or research sector endeavors to raise interest in healthcare and research issues among a particular group in society.
Community involvement (CI) [67]	Sometimes replaced by *community participation*. Often indicates a higher level of involvement than *community engagement*.
Community-based participatory research (CBPR) [67, 90]	A type of research in which representatives from society participate in the research.

Term	Definition
Community-partnered participatory research (CPPR) [115]	A type of research in which representatives from society are partners in the research. Often indicates a higher level of co-creation than *CBPR*.
Community-academic partnerships (CAP) [62, 116]	A type of research environment in which the public are partners in healthcare or in research, often focusing on health-improvement measures.
Comparative effectiveness research (CER) [117]	Research focused on providing patients, next of kin, carers and other stakeholders in the care process with evidence-based information as a basis for health-promotion decisions. Often linked to *PCOR*.
Comprehensive Participatory Planning and Evaluation (CPPE) [118]	A specific type of community-based participatory research with a high level of involvement of representatives from society in the planning and implementation of health-promotion measures.
Experience-based co-design. (EBCD) [35]	A type of *co-creation* in which the experiences of patients or representatives from society form the basis for the development of healthcare or research.
Integrated knowledge translation (IKT) [7]	A type of *co-creation* in which health research results are used in ways that are relevant to all stakeholders.
Patient engagement (PE) [71, 116]	An approach in which healthcare or research organizations seek to raise interest in healthcare and research issues among patients.
Patient involvement (PI) [34, 35]	Used interchangeably with 'patient participation'. Often indicates a higher level of involvement than *patient engagement.*
Patient and public involvement (PPI) [26, 60]	An approach that seeks to involve patients and the public in healthcare and research issues.
Patient-centered outcomes research (PCOR) [86, 116, 119]	A type of individual-centered research with the aim of implementing health-promotion measures.
Patient-oriented research (POR) [120]	Research based on the patient and the patient's best interests.
Power sharing [39, 49, 100, 121]	A type of approach in collaborative settings where participants are aware of power perspectives and actively work to balance the distribution of power among participants.

Term	Definition
Real-world evidence (RWE) och real-world data (RWD) [122]	Real-world data are different types of data collected in real situations in everyday life – this can be seen in relation to the type of data collected in clinical trials. The data forms the basis of real-world evidence, which makes it possible to evaluate how patients respond to, for example, a treatment in their everyday life, outside the controlled environment of a clinical trial. This is an important building block in personalized healthcare. Sometimes RWE/RWD is referred to as 'observational studies' or 'observational data'.
Shared decision making (SDM) [56]	Usually used in healthcare contexts where the patient is actively involved in various healthcare decisions, focusing on both medical evidence and patient preferences.
Stakeholder engagement [115]	An approach that seeks to raise interest in healthcare and research issues among all stakeholders.

What all the above concepts have in common is that they describe an approach that includes experiences and perspectives beyond those of professionals and researchers. The approaches may look different, but they all have the same purpose – to create added value through collaboration.

Appendix 6: Selection of review articles on patient and next-of-kin collaboration

There are several review articles and systematic reviews focusing on patient and next-of-kin collaboration. Listed below are some studies the working group has identified as particularly relevant in this context, including brief summaries of the outcomes.

Title	Summary
Optimizing patient and public involvement (PPI): Identifying its "essential" and "desirable" principles using a systematic review and modified Delphi methodology [26]	A systematic review of 12 systematic studies and 88 occurrences of gray literature, i.e., popular science or other literature not subject to peer review. The results include a ranking of principles that are necessary and desirable to integrate into collaborative work.
Use and reporting of experience-based codesign studies in the healthcare setting: a systematic review [128]	A systematic review of 20 healthcare studies related to experience-based co-design (EBCD). The results show that EBCD can be a useful method for effective co-creation, but that it is important to work according to clear parameters for implementation and reporting.
Frameworks for supporting patient and public involvement in research: Systematic review and co-design pilot [100]	A systematic review of 65 frameworks for collaboration in medical research. The frameworks have been sorted into five categories according to their main objectives. The results show that it can be difficult to transfer a framework directly and that it is therefore beneficial to customize your own solution.
Patient engagement in research: a systematic review [47]	A systematic review of 142 collaborative initiatives in health research. The results show that collaboration is generally possible, but that there is a high risk of initiatives becoming merely symbolic.
A systematic review of evidence on the links between patient experience and clinical safety and effectiveness [123]	A systematic review of 40 collaborative initiatives in primary and secondary care. The results show that collaboration can improve the safety and effectiveness of care.
"Patient participation" and related concepts: A scoping review on their dimensional composition [28]	A scoping review of 39 articles focusing on collaboration in healthcare. The results show that it is valuable to focus on the overall goal of collaboration, rather than trying to differentiate between different types of collaboration.

Title	Summary
Clarifying the degrees, modes, and muddles of "meaningful" patient engagement in health services planning and designing [15]	A systematic review of 18 articles focusing on patient engagement (PE) initiatives in the context of healthcare development. The results show that collaboration benefits from clearly defined goals and other parameters.
Patient engagement in Canada: a scoping review of the 'how' and 'what' of patient engagement in health research [124]	A scoping review of 55 collaborative initiatives in Canada, the US and the UK. The results indicate that better frameworks for collaboration are needed and that a paradigm shift is necessary to normalize patient involvement in research beyond the role of research subjects.
Patients as partners in health research: A scoping review [89]	A scoping review of 119 references from scientific and gray literature. The results show that clearly defined objectives for collaborative initiatives lead to better outcomes.
Community-Based Participatory Research (CBPR) to Enhance Participation of Racial/ Ethnic Minorities in Clinical Trials: A 10-Year Systematic Review [67]	A systematic review focusing on the use of community-based participatory research (CBPR) in clinical trials i nvolving ethnic minorities. The results show that CBPR can facilitate the development of long-term community–academic partnerships (CAP) and that it is a tool for promoting diversity in clinical trials.
Under what circumstances can immigrant patients and healthcare professionals co-produce health? – an interpretive scoping review [125]	A scoping review of 15 articles about co-production or co-creation initiatives in healthcare, with the aim of identifying methods that can facilitate the inclusion of migrant patients. The results show that immigrant patients can be valuable partners in the co-creation of healthcare, but that specific challenges exist for the sustainable inclusion of this minority.
Applying priority-setting frameworks: A review of public and vulnerable populations' participation in health-system priority setting [65]	A scoping review of 96 articles focusing on healthcare prioritization frameworks. The results show that representatives from the public and vulnerable groups were rarely involved, even though the frameworks were developed with the aim of involving all stakeholders.
Engaging patients in de-implementation interventions to reduce low-value clinical care: a systematic review and meta-analysis [126]	A systematic review and meta-analysis of 22 studies focused on using shared decision-making (SDM) to reduce unnecessary or low-value care, mainly in primary care. The results show that SDM is an effective tool for reducing low-value care.

Appendix 7: Frameworks and toolkits

Frameworks and toolkits

Several frameworks and toolkits for collaboration are discussed in the literature. For example, one major study has identified as many as 60 separate frameworks for collaboration in medical research alone.[100] There is no established definition of what actually distinguishes a framework or a toolkit, and the terms are sometimes used inconsistently. The frameworks and toolkits that have been included below contain more general principles for collaboration, rather than those developed for specific contexts. Examples of the latter are Au 2021, where a toolkit for collaboration with patients and next of kin in connection with rounds in intensive care is discussed; Ludwig 2020, where the focus is on collaboration with frail or seriously ill patients; and Mockford 2012, where only studies linked to PPI methodology in the United Kingdom are included. These examples are provided to illustrate the breadth of how frameworks/toolkits might be designed and used.

Framework/toolkit	Summary
A lung cancer research agenda that reflects the diverse perspectives of community stakeholders: process and outcomes of the SEED method [85]	A *Stakeholder Engagement quEstion Development and Prioritization* (SEED) toolkit, which aims to develop research questions that reflect the needs of patients, clinicians and other healthcare stakeholders.
A patient and public involvement (PPI) toolkit for meaningful and flexible involvement in clinical trials [2]	A toolkit focusing on collaboration in clinical trials, in contexts where study leaders are unfamiliar with patient collaboration.
Building Meaningful Patient Engagement in Research Case Study From ADVANCE Clinical Data Research Network [116]	A case study of how the co-created framework ADVANCE has been used in the development of health research.
Co-designed framework to support and sustain patient and family engagement in health-care decision making [88]	A co-created framework focusing on involving patients and next of kin in healthcare decision-making.
Co-production Compass (COCO) [127]	A framework for assessing patient preferences in the context of co-creative collaborative initiatives.
Developing a toolkit for engagement practice: sharing power with communities in priority-setting for global health research projects [121]	A toolkit for initiatives in which researchers and representatives from society work together to jointly define priorities for global research projects with a focus on healthcare.
Enhancement of Comparative Effectiveness esearch (CER) Through Continuous Patient Engagement [117]	A ten-step framework for comparative effectiveness research (CER). Has been applied to several studies focusing on patient-centered outcomes research (PCOR), such as the *PATient-centered Involvement in Evaluating the effectiveNess of TreatmentS* (PATIENTS) program.[86]

Framework/toolkit	Summary
Generative Co-Design Framework for Healthcare Innovation [107]	A framework focused on improving healthcare systems, products and services.
Patient Engagement Translation Table (PETT) [73]	A tool that can be linked to the ten steps of the CER framework listed above. The table links methods for collaboration in research to each step of the framework.
Public Involvement Impact Assessment Framework (PiiAF) [20]	A framework with researchers as the main target group and with a focus on good patient involvement and follow-up of results. Available online.

Appendix 8: Interview guide – experiences of collaboration

These questions were asked in interviews with patient and next-of-kin representatives involved in Collaboration 2.0.

– What are your experiences with collaboration?

– Do you have any good/successful examples of collaboration that you have been involved in?

– Do you have any examples of non-functioning/bad collaboration that you have been involved in?

– Which people/roles are included in collaboration you have been involved in?

– What are your perspectives on collaboration?

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COLLABORATION 2.0:

For a society more sustainable in the long term, where all members are able to contribute and to achieve the best possible health and well-being over time.