D 9.5: SCALING REPORT

Scaling Innovative Care Delivery for Rare diseases & Complex Conditions

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1. INTRODUCTION

Rare diseases are diseases which affect less than 1 out of 2.000 citizens. Nevertheless up to 7% of the European population (or 30 Million people) are affected by rare health conditions (Eurordis 2017). The symptoms of their diseases are often complex, chronical and, due to their rareness, not known (very well) by mainstream health and social care professionals.

As findings of the INNOVCare project have shown, rare diseases affect everyday life of patients and their care givers considerably (Eurordis 2017). They report feelings of unhappiness and depression three times more often than the general population. Since they spend a lot of time on disease-related tasks (care, rehabilitation, medical consultations with different doctors,…) patients and family members have difficulties in following up their professional activities, in completing daily tasks and participating in social life. The burdens they are confronted with derive from the fragmentation of health and social care systems to a high extent. It leads to "a frustrating health and social care experience which often means that individuals are deprived from the best possible care" (Holtgrewe and Gruber 2018, 4).

The project INNOVCare aimed at researching integrated care pathways for rare disease patients, bridging gaps between social and health care and facilitating the organization of everyday life for them and their families. Besides developing and testing holistic care pathways for patients affected by rare diseases in Romania (including one-stop-shop resource centres and case management) the project has investigated possible ways to up-scale these innovations to other European member states and beyond rare diseases (WP9). The following report presents the theoretical and methodological framework of the up-scaling process initiated in WP9. It gives an overview of the analytical framework for the development of the up-scaling model and the roadmaps (D 9.4).

Starting with a literature review, we develop the concept of up-scaling used in the frame of the INNOVCare project. The up-scaling process was initiated in four to three European countries. In the following chapter the authors discuss challenges and advantages of a comparative research perspective applied in the course of the project and focus on the challenges of stakeholder involvement, which is essential for successful up-scaling. In the INNOVCare project expert-interviews and online-discussion

groups were used to explore the different perspectives of relevant stakeholders and to involve them in a discussion with the aim of co-creating a shared vision of integrated care for RD patients.

The analysis focuses on challenges and enablers of scaling integrated care solutions. Thereby three different intervention levels are taken into account: patients' everyday life, the organisation of care and regional and national policies on rare diseases and on the organisation of care. These elements are used for the development of a suitable up-scaling model for INNNOVCARE.

1.1. Concepts of integrated care for rare diseases and complex needs

The needs of people living with rare conditions require coordinated and integrated multiprofessional health and social care. Although good quality health and social care services do exist in many European MS, the existing services often do not meet the needs of patients with a complex condition. This is mostly due to the way those services are organised. Firstly, social and health care services fall into "the responsibility of many sectors, jurisdictions, institutions, and providers" (Kodner & Spreeuwenberg 2002, 3). Secondly, the funding mechanisms and regulations differ heavily among health and social care providers. Thus, care systems are not set up in ways that enable coordination between the two care sectors. Moreover, "health and social care differ distinctively in terms of language, culture, professional roles and responsibilities, and clinical or services approaches" (ibid).

These factors make it very difficult for patients with chronic or multifaceted conditions to access the services they need when they need it. Fragmented care systems make the organisation and coordination of care burdensome for patients living with a rare condition and their family members. In order to increase their quality of life the provision of integrated care should be a priority. This is not just a concern of rare disease patients. Demand for the provision of integrated care is expected to increase across health systems due to demographic change, an ageing population as well as changes in patterns of morbidity (Gröne & Garcia-Barbero 2001), and regional and social disparities (Auschra et. al. 2018). Additionally, the demand for integrated health and social services can be attributed to changes in patients' attitudes and expectations medical services. Patients are becoming more self-confident and are learning to take an active role in the organisation of care. Many countries have strengthened patients' rights. Thus, health and social care systems that move towards integration will not only benefit

patients with rare and complex conditions, but will also contribute to make those systems more sustainable in the future.

On the supply side several developments have contributed to promote the integration of care (ibid). Medical technologies and telemedicine are pushing boundaries of medical care, allowing more services to be delivered remotely. Skill and staff shortages require revisions of labour, for example between doctors and other health professionals. Information technology and digitalisation offer new ways of obtaining and sharing information on patients, diagnoses, and therapies among care professionals of various domains. Finally, since most care systems have come under economic pressure integrated care is seen as a means of delivering care in a more cost-effective way while still ensuring quality and patient-centeredness.

1.1.1. THE SCOPE OF INTEGRATION

Integration of care can mean many things, depending on the definition chosen. The WHO defines integrated care as a form of care that brings "together inputs, delivery, management and organization of services related to diagnosis, treatment, care, rehabilitation and health promotion" (ibid., 7). This definition focuses on a better coordination of the different aspects of health care by the alignment and collaboration of health care institutions and health care professionals. The establishment and mainstreaming of multi-professional teams on the same hierarchical level is referred to as horizontal integration, whereas vertical integration strives for the improved collaboration of the primary, secondary and tertiary care sectors (ibid).

Although the proposed definition of integrated care by the WHO is already far-reaching it does not take social services into consideration. However, the analysis of the multi-faceted needs of rare disease patients (EURORDIS 2017) clearly shows: in order to improve the care outcomes and the life quality of rare disease patients integration has to go beyond the medical domain. In a more encompassing manner integration is then understood as the endeavour "to connect the healthcare system (acute, primary medical, and skilled) with other human service systems (e.g. long-term care, education, and vocational and housing services)" (Leutz 1999, 77f.). The perspective on patients that underlies this

definition does not regard patients as medical cases only, but as human beings fulfilling various social roles and being integrated in diverse social contexts.

1.2. Aims of integrated care

The integration of care is considered beneficial in a two-fold way. First, it is thought to improve the outcomes for patients. This includes the improved access, coordination and continuity of care, more flexibility for patients and care providers and a greater satisfaction of patients and their families. Moreover, the integration of care is expected to improve medical and clinical outcomes as well as the quality of care and in consequence the quality of life of patients and their families (Leutz 1999, Kodner & Spreeuwenberg 2002). On a policy level integration is considered to bring about "greater efficiency and effectiveness" (Kodner & Spreeuwenberg 2002, 2) as well as reducing duplication and the waste of resources (Brown & McCool 1986) within the social and health care system. However, the nature of the relationship between cost-efficiency and integrated care is still under-researched (Hardy et al. 1999, Kodner & Spreeuwenberg 2002, Olsson 2009, Desmedt et al. 2016).

2. UP-SCALING OF CARE INNOVATIONS

Up-scaling describes the effort to **increase the impact** of (social) innovations (Gabriel 2014). This can be done in many different ways. Horizontal or quantitative scaling processes, for example, refer to the extension or expansion of social innovations to render them accessible to a **larger or a new part of the population** (Gabriel 2014, WHO 2009). In the scope of INNOVCare this means to search for potentials to deliver integrated care solutions to more people with rare diseases, disseminating the innovation to new geographical and thematic areas, or to other patients groups with complex needs.

When social innovations, developed on a local or regional level, are to be implemented on a national level and necessary **resources are (re-)distributed** vertical or political up-scaling is taking place. Building on and adding to existing programmes and existing innovations as well as "developing complementary innovations"(Gabriel 2014: 10) is referred to as functional up-scaling. It is addressing the questions how innovations can "iterate, build on and add to social innovations" (ibid., 10). This is also one benefit of

up-scaling social innovations. Building on innovations that have been successfully implemented in other contexts may be more persuasive to risk-averse administrations and policy-makers.

In the context of health and social service innovations the ExpandNet/WHO defines up-scaling more concretely as the "effort to increase the impact of […] service innovations […] so as to benefit more people and to foster policy and programme development on a lasting basis" (Simmons et. al 2007, p. viii). Innovations can improve the quality of care for other target groups (horizontal up-scaling) and the overall quality of the care system (vertical up-scaling).

How can we reach these aims? The first part of the definition refers mainly to a concept of "**growth**". The second part of the definition applied by ExpandNet/WHO address the challenge of **policy implementation.** This is what the dimensions of vertical, functional and political scaling deal with. Hence, in reality horizontal, vertical and functional up-scaling often occur in parallel.

Westley et. al. (2014: 4) characterizes up-scaling processes in general by the tasks to "identifying opportunities and barriers at broad institutional scales, with the goal of changing the system that created the social problem in the first place." Up-scaling here is defined as a form of policy implementation aiming at a regulatory change, structural reforms of social protection systems, also tackling those policies, systems and programs that hinder the further development of social innovations and their implementation. The environmental embeddedness of innovations implies that care innovations, as the one proposed by INNOVcare, on the one hand profit from the framework of the welfare institutions and from links to public institutions. Yet, on the other hand these systems can also hinder the up-scaling of social innovations, due to established divisions of labour and the resulting "silo thinking", organisational inertia, professional closure, and a lack of support and coordination, traditionally provided by the central state (Colombo & Sarius 2017, 87).

Successful up-scaling concepts take the environmental starting conditions into account and build on them (Westley et.al. 2014, 24; Pavlickova 2018). In the case of care service innovations this requires a basic understanding of health policies and related political processes in different regions and countries including "knowledge related to health systems planning, budgetary cycles, financing, programme structures, management, human resources, logistics and information needs"(WHO 2009, 28). This

knowledge on the environmental conditions will allow to adapt the innovation to the "institutional and also cultural and normative environments" (Holtgrewe & Millard 2018, 69) and to make use of existing processes and structures, answering to barriers and obstacles the existing care systems might pose (WHO 2009, 27). Moreover knowledge on the different stakeholders and on their social environment is essential for up-scaling processes. Health care organisations usually comprise very specialized entities that differ in their working style, their means, their focus and as a result, the perspective through which they assess the patient or the condition. Hence, for the successful implementation of integrated care it is crucial to develop clear and shared objectives (see West & Poulton 1997).

Stakeholders can tell us about the circumstances we will find in a region, country or a political field and are relevant partners to develop the social innovation in a way which is most profitable for its environment (WHO 2010, 11). Building scaling processes as negotiation processes allows engaging several different actors with different skills, knowledge, and background and to develop a more encompassing view of the problem. The participation of relevant key-players is therefore considered as the first and most important step to start up-scaling processes (WHO 2010).

However, since experiences and perspectives of stakeholders often differ strongly, up-scaling is complex and difficult. Stakeholders' aims can interfere with the overall aim of the social innovation delivered. Including them in the design of the up-scaling process helps to establish of a shared strategy and clear and ambitious goals (WHO 2010) and can foster personal involvement of the relevant key players. Hence, up-scaling processes and the social innovation delivered inevitably are products of the negotiation process between relevant stakeholders and interest groups and require considerable "institutional work" by promotors and their allies (Windrum et al. 2018). Both result from multiple interrelated actions, modes of learning, conflicts and tensions which derive from cooperation and compromise between stakeholders. These can have intended and unintended consequences (Holtgrewe & Millard 2018: 70). Up-scaling has therefore also been described as a "collaborative innovation that occurs between the actors and stakeholders that populate [the innovations] environment" (Terstriep et. al. 2015, 96). The ways in which stakeholders can get involved are manifold: Social innovations aiming at up-scaling can spread information, build committees, partnerships, affiliations (Benisi 2016) or intra-organizational relations (Terstriep et. al. 2015, 87).

Having the different stakeholders in mind, different directions and scopes of up-scaling as well as strategies how to scale up social innovations can be formulated. In a bottom-up process initiated by patient organizations or care givers, community based solutions play a significant role (Westley et.al. 2014, 17). They facilitate the emergence of new local networks and partnerships, building on existing community assets and empowering people. Contrary to that, a top-down process starts distribution of innovation from the policy level.

Yet, in the area of public policies up-scaling processes seldom follow only one strategy. As Holtgrewe and Millard (2018, 69) argue "a double-pronged strategy in which bottom-up approaches simultaneously solve problems and develop the agency of social innovators and beneficiaries, whilst top-down approaches create a supportive political and regulatory frameworks and also mind-sets and ways of living and working" is often most successful. Successful scaling can therefore not be developed in one direction only but has to address different levels of intervention simultaneously.

Summing up what has been said in the last chapter, the following aspects need to be taken into consideration when searching for up-scaling possibilities and strategies of innovative care models:

1) Successful scaling needs to consider how the social innovation delivered can **adapt to the institutional, cultural and normative environments** it is both improving and challenging. Hence we need to develop a basic understanding of the organizational, national and regional circumstances, different care systems and the challenges they pose for rare disease patients and their families. They are a starting point for the development of an up-scaling vision.

   Successful up-scaling processes should seek to establish relationships with as many relevant stakeholders as possible. Stakeholders' different perspectives and positions in the field of rare diseases need to be taken into account. Up-scaling processes should seek to involve stakeholders of all levels – policy makers, care professionals, patients and patients' representatives. The involvement of policy makers is essential to enable implementation.

   When we know who is to be involved in the up-scaling process we can decide how the care innovation delivered can be scaled up. It is favourable to involve stakeholders already in the conceptualization of the up-scaling process. This process of co-creation can foster the development

a **shared vision** of the problem and helps to develop common aims. Moreover it can strengthen the personal commitment of stakeholders.

2) As the innovation delivered can be linked with "a diversity of goals and take different meanings over time, depending on the wider political concept and institutional system" in which it is embedded, the innovative instrument (e.g. case management) can work and be judged differently in different places and circumstances (Evers et. al. 2014: 11). Up-scaling therefore also implies a change of the innovation itself, including possible unintended consequences and ways of addressing them.

   The direction that up-scaling processes take, their outcomes and the strategies followed depend on the target groups of up-scaling, the social, political and economic environment they are embedded in, the problems addressed and the stakeholders that are involved. Indeed, such processes are the result of their real-life interplay. Up-scaling can therefore not be considered as linear model, linking policy directly to its implementation (WHO 2009), but rather has to be considered as a "circular" "adaptive" or "evolutionary" concept (Ernest R. Alexander 1985, 1). Outcomes of up-scaling process are therefore never definite. However, when this uncertainty and open-endedness is accepted and even embraced, there are possible gains also in a processual perspective: here up-scaling activities can build stronger relations between important stakeholders, creating platforms for exchange and for the development of further tools and concepts to approach integrated care for people with rare diseases.

3. Methodological approach

In the previous chapters we have argued that up-scaling strategies deal with two decisive questions:

1) How can social innovations be adapted to the environmental circumstances?
2) Who needs to be involved in the up-scaling process?

These questions structure the following chapter consisting of three main sections. First it will discuss the challenges of identifying opportunities and obstacles to adapt care innovations to the environmental circumstances. To identify obstacles and opportunities for up-scaling the authors are following a cross-national comparative research approach. Secondly, the chapter will explain the

approach to stakeholder involvement and the way experts of the field got involved in the INNOVCare up-scaling process. Finally, the analysis of expert interviews and of the role of expert-meetings is explained.

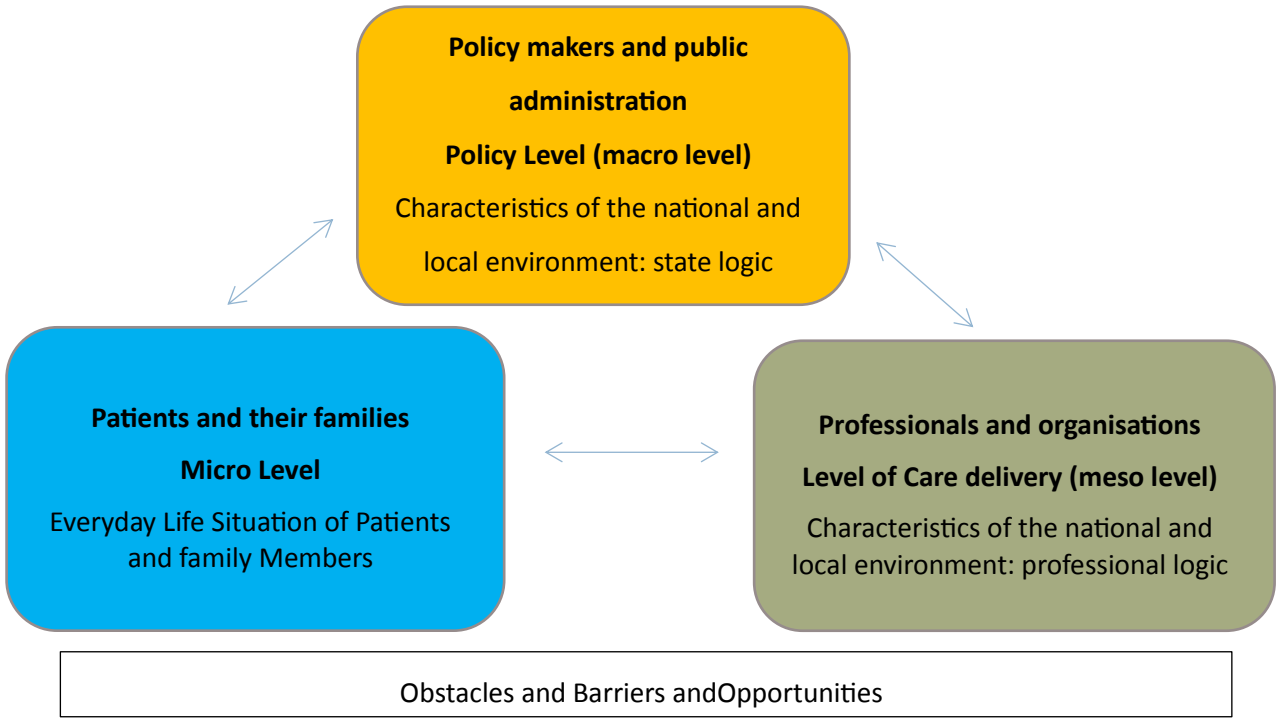
3.1. Adapting innovations to the environment circumstances

Care systems are complex and fragmented. Legal frameworks and processes of decision making, professional logics, welfare traditions and other characteristics of the national and local environments influence the everyday life of rare disease patients and their families as well as organisational and professional care delivery and the policies shaping care systems. What we have called as environmental circumstances or contexts in the previous chapter can also be characterised as the landscape of care, structured by different possible levels of intervention:

- The policy level (the macro level), which is operating according to a state's logic,
- The level of care organisations or the professional sphere (the meso level) which involves different stakeholders and interest groups who are interacting with the stakeholders of the other levels,
- and patients and their relatives, as the one receiving care and participating in the design of its own care (the micro level)

These intervention levels are guiding the identification of possible next steps, sub-aims and scaleable domains. All three levels of intervention are connected and hierarchically linked. Moreover, each level of intervention is characterised by different operational logics and decision making processes involving stakeholders from different domains and levels. In the scope of INNOVCare we are focusing on the relations between the different levels of intervention, thereby asking how a stronger coordination between stakeholders of these different fields can be fostered, which barriers need to be overcome and in how far operational logics need to be changed.

FIGURE 1. INTERRELATED LEVELS OF INTERVENTION AND THEIR STAKEHOLDERS



The guiding principle for up-scaling processes on a micro level is the individual benefiting from social innovations (Holtgrewe and Millard 2018, 71). As holistic care models always put the needs of patients in the centre (WHO 2009, 29), the micro-level – i.e. everyday life situations of patients and their families - can be considered as one important starting point to design up-scaling processes. For integrated care to really reach its potential the involvement of patient and family (carers), on the micro-level, as well as further research and evaluation of the integration efforts are paramount (Kodner & Spreeuwenberg 2002). Analysing the micro level means to focus on the immediate impact of rare conditions and the way care is organised on everyday life. This is what the research done in WP4 of the INNOVCare project has focused on and what has been implemented and tested in NORO, Romania (WP6 and WP7), providing a case management based approach to integrated care for rare disease patients and their families.

The meso-level is addressing the organisational level of care institutions and professionals, including also interest groups and non-governmental organisations. In the scope of INNOVCare these are for

example hospitals, care service providers, general practitioners, schools, transport providers, professional associations and patient organisations. The integration of care across or even within health care organisations is a highly complex endeavour, since those organisations count among the most complex ones. On the meso level the professional logics of different stakeholders need to be taken into account. The aim of exchange between stakeholders of different profession is to establish a common understanding of integrated care. Here "differing rules, inter-sectoral boundaries (as between health care, mental health care, and social care), funding streams, and institutional and professional cultures" (Kodner & Spreeuwenberg 2002, 2) pose potential obstacles to the integration of care.

Interventions on the macro level – the policy level – have the possibility of tweaking the structures of care. The macro level, as the one influencing the way the care system is set up, is the most important enabler or hinderer of change and yet the one which is changing only very slowly. On the macro level the separation of political domains and responsibilities and legal frameworks are one of the main challenges for integrated care solutions.

3.1.1 Comparative research in the field of social innovation

The up-scaling activities of INNOVCare took place in four different European member states. Analysing up-scaling possibilities in four different countries we aimed at the identification of similarities and differences between enablers and barriers to integrated care. This comparative approach allows building knowledge on how social innovations contribute to innovative care solutions in different contexts. It enables us to detect local peculiarities, similarities, advantages, disadvantages and innovative solutions to encounter obstacles. This way the comparison can inspire new and innovative solutions. Moreover, it fosters a European perspective as problems and enablers can be analysed across cases (Moulaert & Mehmood 2013, 444). This also answers to the special situation of patients with complex and/ or chronic conditions and their families that is, despite the differences between care systems, quite similar in different European countries (EURORDIS 2017). Comparing different settings of up-scaling processes is then becoming a "part of the social learning process involved in many social actions and movements" which is "informing and enhancing collective action and public policy" (Moulaert & Mehmood 2013, 442).

By deciding on a comparative perspective we set aside an in depth analysis of the national care systems. Instead of an encompassing analysis of the national systems, reviewing regional particularities systematically, we choose some best-practice examples of communities and regions in all four countries. In addition, stakeholder contacts that were established in the course of the up-scaling process guided the selection of communities.

3.1.2. COUNTRY SELECTION

Both health and social care systems, which are in the focus of INNOVCare project, are highly varied by country and also region. They have developed historically, in specific configurations of welfare policies. Stakeholders and their knowledge bases (for example, different professions), norms and interests influence the systems functions. INNOVCare took a comparative approach for which a heterogeneous sample of countries was selected. The following criteria were taken into account: political strategies and policies toward people with rare diseases, the type of welfare states and public administration, the size of the country and the population, GDP per Capita and their geographic representation across the EU. Also feasibility was an important criterion for the country selection, enabling a stronger involvement of different stakeholders, who had been involved in the project before. Based on this criteria Austria, Romania, Spain and Sweden were selected for the up-scaling process.

FIGURE 2. WP4-CRETERIA FOR THE COUNTRY SELECTION

Diversity	Policy	Feasibility
✓ Type of welfare states ✓ Size of the country ✓ Size of the population ✓ Type of public administration ✓ GDP per Capita ✓ Geographic representation across the EU	✓ National plan for rare diseases (RD) ✓ Reference to social issues in national plan for RD and in RD national multi-stakeholder conference ✓ Resource centres for RD ✓ Pilot for case management	✓ Country of INNOVCare partners ✓ Existence of National Alliances of RD patient organisations ✓ Involvement in similar EU projects

The heterogeneous sample allows identifying a variety of potential obstacles regarding integrated care as well as concepts and possible ways to approach them.

To distinguish between different welfare state traditions we drew on the typology of Esping-Andersen (1990). Esping-Andersen categorises welfare states based on the degree of de-commodification, i.e. the

extent to which welfare policies free individuals from market dependency, and the particular welfare mix between the market, the state and families. In this regard, Austria can be characterised as conservative-corporatist welfare state, which has a strong focus on employment-related social insurances and a federal system of care provision. Sweden is characterises as a social democratic/ universalist welfare state. It provides access to benefits and services based on citizenship, extending social insurance to cover the whole population instead of compensating wage earners (and their relatives) (Anneli Anttonen & Jorma Sipilä 2015). Spain as a South European welfare state is not well covered by the classic "worlds of welfare capitalism" of Esping Andersen. On one hand the system has strong elements of a conservative-corporatist welfare state, but one the other hand the health care system is based on universal entitlements for all citizens irrespective of their labour market status (Ferrara 1996). Romania's welfare system can be characterised as a rudimentary welfare regime in transition or as a post-communist welfare regime. It struggles with the challenges to meet the rising social costs of transition to a market system (Sotiropoulo 2003, 658), shifting responsibilities from a national to a regional level and cutting costs by austerity programs (Pop-Radu 2014, 175). Partial privatisation deteriorated public health services and worsened service provision for the poorest parts of the population (Sotiropoulo 2003, 665) Also regional differences in Romania and the generally scarce financial resources are a challenge for the health and social care system.

Furthermore, we drew on feminist perspectives on welfare states, which are highly relevant in the context of health and social care, because they pay particular attention to the gendered division of paid and unpaid labour as well as the provision of informal and formal care (Sainsbury 2008). Thus, they expand Esping-Andersen's' typology by highlighting how care work is shared within a certain regime. The welfare regimes of Spain and Austria are organised along the male breadwinner-model. Men are the primary wage earners while women perform the predominant portion of care work (Lewis 1992). Hence, gaps in professional provision of and health social care tend to be complemented or compensated to a large extent by family-provided care in these countries. In Romania with its limited services, care is mostly provided by female relatives. Hence women carry the double burden of paid work and care work. Often they reduce their paid employment with consequent losses in income and social security. However, through social change, increasing labour market activity of women, regional

mobility and, especially in Romania, emigration of both women and healthcare staff, the role of women and families in complementing conservative or patchy social services is coming under pressure. Some welfare state theorists speak of a "reproduction crisis" in both market and non-market segments of care (Aulenbacher 2011).

In Sweden formal care is well developed and this includes care delivery to people with rare diseases. The services respond to the needs of people with complex diseases, offering varied, specified possible ways to make care accessible to the persons. The main goal is to foster the social participation of people with all kinds of special needs. Sweden in the context of our report serves as a comparative case, while we focus on Spain, Austria and Romania. This allows showing the added value integrated care approaches can deliver in different national contexts and the different degrees of institutionalization of such concepts.

3.2. Stakeholder involvement and expert knowledge production

Involving "the right persons" who are committed to engage in further up-scaling process is essential for successful up-scaling. These relevant persons can be referred to as experts. To get to know the experts and activists who need to be involved in the up-scaling process researchers propose to focus on the negotiation processes and in particular the patterns of establishing consensus as well as differences between options (Meuser & Nagel 2009, 27).

In the scope of INNOVcare we addressed the challenge of stakeholder involvement in two ways. First we conducted expert interviews. The expert interviews allowed us to establish contact with the relevant stakeholders and to involve them in the conceptualization of a possible up-scaling strategy. Moreover we gained further inside into the initial starting conditions in each country, detecting existing competences and resources, obstacles and opportunities that had to be considered in the up-scaling process. Additional literature reviews on the health and social care systems helped us to find out about barriers and opportunities of integrated care in the different national settings.

Secondly we organised joint online-discussion groups with the experts to present them preliminary result extracted from the interviews. These discussion groups aim at the establishment a shared vision

and common goals to be reached in the respective national contexts. The discussions helped to gather more in-depth information about the necessities and requirements to implement integrated care projects. They serve as a starting point for the formulation of the possible roadmaps to integrated care, presented in chapter 5.

In addition, the project, drawing on existing contacts and the networking activities of EURORDIS, conducted three workshops on integrated care involving patent organisations, policy contacts and liaising with both the more medical-oriented European Resource Networks and the integrated care community. Participatory discussions of policy and the role of patient organisations gave further insight and contextual information.

3.2.1. EXPERT INTERVIEWS

Expert knowledge is not only professional knowledge. Following Meuser and Nagel (2009, 24) individuals who have acquired their "knowledge on a particular problem through an activity which is aimed at the problem and, therefore, with a view to analysing and or helping to solve the problem" can also be perceived as experts. In expert interview we drive to collect information of action strategies and criteria of decision-making connected with a particular position (Meuser and Nagel 2009, 32). This conceptualization of expert knowledge is for example, referring to members of citizens' groups, self-help groups or NGO'S playing an important role in shaping the care situation of people with rare diseases. Their expertise "is socially institutionalized and linked to a specific context and its functional requirements" as Meuser and Nagel (2009, 24) argue. This broad definition of experts and expert knowledge highlights the specific function such individuals have with regard to the societal configuration of a social problem. Following this argument, experts able to inform on the organization of care for people with rare diseases and complex needs are not only policy makers from the social, the health and the educational field, representing regional, local or national institutions or health and social care professionals but also patient representatives, patients and their care-giving relatives – especially those that have taken own steps of institutionalising their perspective by engaging in self-help groups.

As representatives of civil society they are influencing the way problems are negotiated. Yet, their different institutional backgrounds determine their possibility of participating in the communicative practices and negotiations of problems, strategies, options and solutions (Meuser & Nagel 2009, 31).

They do not only possess a different level of information, but they also take a specific role in the production of expert knowledge and the configuration of the problem. To give some examples: Regional policy makers responsible for care might have a broad institutional knowledge on services provided, but little information on the needs of rare disease patients and their families. Health policy makers tend to focus on the health professions and institutions, often forgetting the "social-life dimension". Policy makers from the social ministries might see the needs of patients with rare diseases as solely health-related and thus do not consider themselves as responsible or they only see the long-term care component while forgetting about other health and life related needs. Rare diseases patients, their relatives and patients organizations thus frequently find that they and their needs fall between the responsibilities of health and social services and established negotiations of policymakers and health care providers. Additionally, the division of decision-making power between regional and national authorities influences the way stakeholders conceive their possibilities to engage in projects achieving patient-centred care. The same is true for care professionals: In many countries the exchange of expertise between health and social care specialists is not well established. Primary care institutions or primary care doctors are often the first contact point for rare disease patients. Yet, they might have little time to deal with the complex needs of patients and little knowledge on their specific needs. Moreover the education sector is often not included in the organization of care provision although it plays a crucial part in ensuring social inclusion.

3.2.3. SELECTION OF INTERVIEW PARTNERS

As up-scaling processes need to involve different stakeholders from different levels we selected interview partners of all different institutional levels asking them how they envision patient-centred, holistic care and which potentials they see to improve the social and health care provision for people with rare diseases/ complex needs in their country and context.

For the expert interviews we selected experts who are taking part in health and social policy making processes as well as care giving professionals involved in integrated care projects. Hence, we interviewed national and regional politicians as well as public administrators of the health and social

sector, service providers, case managers and patient representatives, include different experiences and perspectives of relevant stakeholders.

We interviewed experts who had already been involved in the INNOVCare project, carrying on the discourse developed in the project in order to strengthen the established relations profitable for the up-scaling. Building on their networks we aimed to involve further interview partners ("snowball sampling"), who were considered as important partners by the experts interviewed. Further criterion for the selection of interview partners were their decision power and their expertise in the relevant policy fields. Therefore we aimed to reach out to policy makers so far not involved in the project. In total, we interviewed 34 experts in four countries, listed in the table below. Experts who have been interviewed together are not listed separately.

TABLE 1: STAKEHOLDERS INTERVIEWED

Expert	Current position	Expertise on RD	Country
E_1	Public administration	National Agency for Rare Diseases, Ministry of health	Austria
E_2	Two Public administrators	Care and health, regional authorities, involved in a project on community nurses	Austria
E_3	Consultant	Member of the Accreditation Council of ERN's; responsible for the designee of curricular for care givers and other health professionals	Austria
E_4	Policy maker	Ministry of labour, social affairs, health and consumer protection	Austria
E_5	Patient representative	Co-founder of a resource centre, representative of the umbrella organization of RD patients	Austria

E_6	Service provider	Competence Centre for integrated care	Austria
E_7	Representative of a patients organization	Responsible for an information hotline for RD patients and their families	Spain
E_8	Representative of a patients organization	Delegate of an autonomous region; coordinator of the Spanish clinical group of a certain RD, trains other professionals on the subject of RD	Spain
E_9	Representative of a patients organization	Association offering training and aiming at the empowerment of patients	Spain
E_10	Case manager	Case manager working with children with rare disease and severe disabilities	Spain
E_11	Case manager	Case manager working with patients and families confronted with a specific RD	Spain
E_12	Policy maker	Social services; regional authorities	Spain
E_13	Two Policy makers	Ministry of Health, Social Services and Equality, Policymaker responsible for the formulation of the national strategy on RD	Spain
E_14	Two Public administration officers	Health services; regional authorities	Spain
E_15	Policy maker and public administration officer	Health services; regional authorities	Spain
E_16	Consultant	Doctor, consultant for the ministry of health on RD	Romania
E_17	Policy maker	Ministry of health	Romania

E_18	Policy maker	Public Health Directorate	Romania
E_19	Policy maker	Ministry of labour and social justice	Romania
E_20	Policy maker	General Directorate for Social Assistance and Child Protection, regional authority	Romania
E_21	Policy maker	Councillor, City hall	Romania
E_22	*Four Case managers*	*NORO*	*Romania*
E_23	Patient representative and social service provider	Co-founder of a resource centre	Romania
E_24	Policy maker	Social services, local authorities	Romania
E_25	Consultant	Doctor, employed in a Centre of Expertise; consultant for the ministry of health	Romania
E_26	Service Provider	Co-founder of a resource centre for RD patients	Sweden
E_27	National focal point for RD	National Agency for RD, founded by a National body under the Ministry of Health and Social Affairs	Sweden

In **Spain** we interviewed twelve experts: Three representatives of patient organizations, two case managers, one care provider and six health and social care policy makers/public administration officers of different regions and of the national level. We selected interview partners from national bodies and from Murcia, Catalonia (dominantly Barcelona) and Andalusia; firstly, because policymakers of these two regions were already involved in the INNOVCare project. Secondly, because these two regions can be considered as best practice examples in the provision of health and social care for people with rare diseases. In Murcia a new care plan for people with rare diseases has been developed, involving all

relevant stakeholders. In Catalonia case management is been implemented for people with rare diseases at the hospitals. As case management is organised differently in Catalonia than in the INNOVCare model tested in Romania, the Catalonian case was very interesting as a comparative care in INNOVCare, showing different possibilities of approaching and implementing professionalised care coordination. Moreover, we conducted an online-interview with patient's representatives in Andalusia, which was named by FEDER as a province facing more difficulties.

In **Austria** we conducted seven interviews. We interviewed five health and social policy makers, one representative of the umbrella organization of patient organizations, representing also a centre of expertise and one representative of the competence centre for integrated care. Besides national policy makers we interviewed regional policy makers from Carinthia, who took part in an EU-funded pilot project, which aims to improve the situation of chronically ill patients[1] and is employing family and community nurses. In addition, we included information collected during the 8th Austrian Congress for Rare Diseases conference organized by the patient representative in the analysis as well as answers on the parliamentary question on rare diseases in Austria.

In **Romania** we conducted in total ten interviews: Seven health and social policy makers, four case managers and two service providers of whom one was also speaking as a patient representative, were interviewed. Besides national policy makers, we interviewed experts from Salaj County, where the NORO-centre is situated and from Botosani, a region with little resources and structural problems in setting up integrated health and social care systems in the east of Romania, where community nurses have been installed.

In Sweden we built on the existing contact to Ågrenska, a national competence centre which provides programs for people with rare diseases and their families, to find interview partners. As the community is very well connected in Sweden, building on a long tradition of cooperation on different levels, is serves as a best practice example in our project. Hence, we will not develop a national roadmap for

[1] http://www.renewinghealth.eu/

Sweden, but take the information collected in Sweden into account, when developing potential up-scaling roadmaps for Austria, Spain and Romania.

3.2.4. ANALYSIS OF THE EXPERT INTERVIEWS

The expert interviews were analysed by conducting a qualitative content analysis, focusing on the topics and thematic units addressed by the experts. Codes were first built in a deductive way, using literature on integrated care and up-scaling. Then they were complemented by inductive categories taken from the data. Expert interviews where not fully transcribed. Instead notes taken during the interview were used as basic information. They were complemented with additional information of thematically relevant passages.

Codes are organized according to steps that need to be taken in an up-scaling process. Thereby the structure of the care systems and of possibly levels of intervention on one hand, and the possible interactions between the different levels on the other are taken into account. They are not only guiding the definition of possible codes but also the formulation of recommendations, identifying possible intervention areas and next steps, identifying sub-aims on the different levels of intervention. (Gruber & Holtgrewe 2017).

The main categories used for the analyses of the expert interviews are:

1) Characteristics of the stakeholders involved:
   - The experts position in the field of (integrated) care and rare diseases
   - Stakeholders named that need to be involved in an up-scaling process
2) Policies on rare diseases – the macro level:
   - Characteristics of the national and local environment following the state logic
   - Policies on rare diseases
   - Strategic aims
3) Care delivery – the meso level:
   - Characteristics of the national and local environment following the professional logic
   - Tools of integrated care

- Patients involvement

4) As crosscutting topics concerning all three levels:
   - Obstacles and barriers
   - Opportunities
   - Elements of integrated care

Each of these main categories contains a certain number of sub-categories. On the whole the coding system contains 142 codes, capturing each three different sub-levels of codes.

1) Characteristics of the stakeholders involved:

We have differentiated between four possible **expert positions** in the field of rare diseases and integrated care: (1) representatives of official institutions, who have expert knowledge on integrated care, (2) representatives of patients' organizations and non-governmental organizations, (3) professionals and service providers (including doctors, case managers and nurses), (4) regional and national policy makers with expertise and decision making power in the social, health and/or educational field.

S**takeholders that were named by the experts as important players in the field** were coded separately. Here, the division in different levels of intervention helped again, to identify stakeholders and codes. We differentiated between patients (micro), stakeholders and organizations involved in the organization of care and structuring everyday life (meso) and policy institutions (macro). Organisations delivering care are divided into mainstream organizations and specialized services for rare disease patients. Mainstream organizations are besides care institutions, such as hospital, the social insurance funds, and primary health care centres of family doctors, the working place and schools. Specialized organizations are the European network of reference centres, centres of expertise and resource centres for rare disease patients, the pharma industries producing orphan drugs and case managers. On the policy level we differentiated between the different policy levels (local, regional, national and European level) and the different stakeholders involved, i.e. administrative staff of public institutions, policy maker, patients' representatives, and external experts, representatives of non-governmental organizations, and parliamentary groups or parties.

In order to capture the **characteristics of the national and local environment** the experts are embedded in, topics experts attributed to the organization of their local, regional or national care system were coded separately. They can be divided according to the logic they are following on a meso or macro level of intervention.

2) Policy level (macro level):

Strategic aims experts mentioned in the interviews allow identifying possible common action strategies and opinions of the relevant stakeholders involved. In our analysis action strategies are differentiated according to the problems they aim to solve. These are, in our case, the links between different stakeholders (including network building, cooperation and political boards), accessibility of care services, the empowerment of patients, and strategic foci in the organization of care (continuity of care, maintenance of health and holistic care visions).

The codes on **policies on rare diseases** sum up information on institutional frameworks and the political scope of policies on Rds. They give insight into the way of policy implementation in the field of integrated care and – in a comparative way – into the regional and national systems characteristics. Codes divide between different strategies of political planning (adapting new services to existing resources, establishing special offers for RD patients and their families, building up local and regional expertise on rare diseases, monitoring tools), legal frameworks influencing the care situation of people with rare diseases(regulation of the supply of patients and the control of the use of resources), political responsibilities (including the establishment of a special department dealing with the needs of RD patients) as well as recommendations and frameworks (National action plans and their implementation as well as the role of key policy makers) important for the further development of rare disease policies.

3) Care delivery (meso level):

The level of care delivery is capturing the characteristics of the national and local environment for the logic of the professions involved. Moreover, it captures t**ool of integrated care**. These tools include for example protocols of treatments, the establishment of multidisciplinary teams and case management (and the different functions and duties assigned to case management). Besides, knowledge building on rare diseases was coded as an element of integrated care. It includes education of professionals and

patients on diseases and services, research on rare diseases and integrated care and training of professionals, patients and main care givers. Besides resources of social care, sectors which have to be taken into consideration in order to establish integrated, holistic care systems have been coded. These are schools and education facilities, employers, providers of support for care givers to organize everyday life, financial transfers, call lines and psychological support for patients and their families. Moreover, human resources named by the experts that are needed to organize integrated care have been collected as separate codes, including psychologist, therapists, mobility support, teachers, nurses and doctors.

4) Cross cutting topics:

Obstacles and barriers are coded according to the level on which they were observed by the experts, i.e. the micro- meso- and macro level of possible interventions. The micro-level is capturing the problems patients with rare diseases or complex needs and their families encounter on an individual or household level. Codes range from psychological burdens, a lack of mobility, and lack of information, stigmatization and financial burdens that could be resolved or mitigated, to personal competences of rare disease patients that could be built up by implementing integrated care solutions, such as their ability to accept help, to organize for their interests and to gain confidence in their knowledge on their diseases, which enables them to organize their care situation accordingly.

The meso-level is capturing problems and obstacles observed by experts on the level of health and social care delivery, such as the lack of coordination between different services, doctor-centred care systems, a lack of services, problems in providing continuity of care and missing competences of professionals to take the expertise of patients into account.

The macro level is capturing obstacles in the field of policies, such as a lack of financial resources for services and drugs, legal frameworks, the division of power between policy makers and a lack of awareness for the needs of patients with rare diseases among policy makers and the lay public.

Opportunities refer to models and projects of integrated care that could also work for rare disease patients, which are already implemented in the different member states selected for the up-scaling

process. Moreover the potentials of the INNOVCare approach to benefit other patient groups (such as patients with multimorbidity or patients with chronic diseases) have ben coded under this domain.

3.2.2. EXPERT (ONLINE) DISCUSSIONS

After the expert interviews were conducted and analysed we held a second round of expert consultations that had three aims. Firstly, we wanted to bring together experts and policy makers in the field of integrated care. Therefore, we organised (online) discussions to facilitate their engagement and their exchange. Secondly, these discussions contributed to validate of our findings from the expert interviews. Thirdly, based on the findings the discussions aimed at identifying possible next steps that need to be taken to facilitate integrated care in the three countries in questions. Thus, the results of the expert discussions form the basis of the road-maps and constitute an integral part of the scaling report.

The participants of those discussions consisted of two groups, experts who were previously interviewed and experts who were not involved in the process so far. This composition enabled us to fulfil all three aims of the (online) discussions, namely the exchange of ideas and positions, the validation and the discussion of possible next steps. The following table specifies the participants of the (online) discussions that were conducted in all the three countries chosen for the upscaling process.

TABLE 2: STAKEHOLDERS INVOLVED IN THE (ONLINE)-DISCUSSIONS

Country	Experts involved
Austria	• Public administrator, Care and health, regional authorities, involved in a project on community nurses • Consultant, Member of the Accreditation Council of ERN's; responsible for the designee of curricular for care givers and other health professionals • Policy maker, Ministry of labour, social affairs, health and consumer protection • representative of the umbrella organization of RD patients

	• Service provider, Competence Centre for integrated care
Romania	• Two patient representatives and social service providers, Co-founder of a resource centre • Consultant, Doctor, consultant for the ministry of health on RD • Law adviser National Authority for Disabled People, The Superior Commission • Policy maker, Ministry of Work, Family, Social Protection and Elderly • Policy maker, Ministry of health • Policy maker, Public Health Directorate • Policy Maker, City Manager • Social services, local authorities local social service provider
Spain	• Representative of a patients organization, Responsible for an information hotline for RD patients and their families • Representative of a patients organization, Association offering training and aiming at the empowerment of patients • Two Policy makers, Ministry of Health, Social Services and Equality, Policymaker responsible for the formulation of the national strategy on RD • Public administration officer, Health services; regional authorities • Policy maker and public administration officer, Health services; regional authorities

Due to the comparatively low level of regional dispersion of participants it was possible to organise the expert discussions as a physical meeting in Austria. However, in Spain and Romania physical meetings bringing together all the stakeholders in one place was not feasible. Therefore the discussions those two countries were held online.

Before the (online) expert meetings the participants received a working document characterised the national health care system as a starting point and that summarised the most important findings along the following dimensions: the concept and aims of integrated care, context-specific obstacles to

implement integrated care and possible starting points and tools for integrated care. Based on this document the following questions were addressed in the meetings.

- Are the obstacles identified shared by the participants and to what extent?
- How can the obstacles identified be overcome?
- What are the means to overcome these obstacles?
- Who needs to be involved to solve these issues?
- In which of the areas of integrated care do measures seem most promising to improve the situation of rare disease patients and their families?
- Which are the short-term, medium-term or long-term goals in the context of the obstacles identified?

In Austria and Romania these questions were discussed during one meeting whereas in Spain two meetings took part to cover all the questions.

The results of the expert discussions form an integral part of the analysis. They were used to refine the working documents and formulate the roadmaps for the countries.

4. Challenges and enablers of scaling innovative care solutions

4.1. CHARACTERISTICS OF THE SELECTED COUNTRIES

The following chapter deals with some characteristics of the care systems in the four countries selected for the up-scaling process. Additional literature has been taken into account for the characterisation of welfare traditions and care systems. Moreover it focuses on rare disease policies as described in the recently published country reports in the frame of the project rare disease -action.[2] We are thereby focusing on the coordination of social and health care, as one of the main challenges addressed by INNOVCare. Hence, other issues related to RD such as research, orphan drugs and diagnoses, which are

[2] http://www.Rd-action.eu/rare-disease-policies-in-europe/

essential for rare disease patients, are only touched upon, but not explained in detail[3]. Instead, we focus on the questions where the issues of policy, care delivery and patient empowerment converge.

TABLE 3: CHARACTERISTICS OF SELECTED COUNTRIES

	DIVERSITY					POLICY		
	Welfare State	**Public administration**	**contry size**	**population**	**GDP per capita/2017**	**NAP**	**RC**	**Pilot CM**
Austria	Conservative/ Corporatist	Regional / Federation	medium	medium	high	YES	YES	NO
Romania	Eastern European	Unitary / Decentralised	medium	medium	low	YES	YES	YES
Spain	Conservative/ Corporatist	Regional / Autonomy	large	large	medium	YES	YES	YES
Sweden	Universalist	Unitary / Decentralised	large	medium	high	NO	YES	NO

Country size: "L (>240 000 km2)", "M (between 50 000 m2 and 240 000 km2" "S(<50 000 m2)" Population size: "L (> 20 000 000 hab.)", "M (4 000 000 to 20 000 000 hab.)", "S(<4 000 000 hab.)"; GPD per capita/Euro 2014: "High(> 110)", "Medium(80 to 110)", "Low(< 80)" [4]; NAP= National Plan, RC=Resource centre, CM=case management (see memo of WP4)

4.1.1. ORGANISATION AND RESPONSIBILITIES FOR THE PROVISION OF CARE AND CARE DELIVERY

Health and social care systems in all the compared countries are decentralised, albeit to a varying extent.

In **Spain** health and social care are devolved to the regional level of the autonomous regions, which also decide on health expenditure. The main role of the national Ministry of Health and Social Policy (MSPS) is to ensure equity between the autonomous regions by setting minimum thresholds for services and expenditures. In addition, the ministry is coordinating the policies of the 17 autonomous regions that

[3] An in-depth analysis of these topics can be found in the country reports of the above named project rare disease -action: http://www.Rd-action.eu/rare-disease-policies-in-europe/

[4] GDP per capita is used as an indicator for the resources countries have – not taking into account, how the resources are distributed in the countries. Of the four selected countries Sweden (Rank 7 of 45 Ranks) and Austria (Rank 10 of 45 Ranks) are ranging relatively high compared to other European counties, while Spain is ranked in the middle (Rank 17 of 45 Ranks) and Romania (Rank 31 of 45 Ranks) relatively low. http://www.imf.org/, dl. 21.6.2017

are accountable only to the regional parliaments and thus not hierarchically linked to the national level (García-Armesto et. al 2010, 38). The legal base for the coordination of care is the Spanish National Health Service (SNS) Cohesion and Quality Act (16/2003). It is also the basis for designating reference centres, departments and units (RCDUs) as it sets out the legal framework for coordination and cooperation between public health authorities in the exercise of their respective functions and defines reference services that require the centralization of cases in a small number of centres. This is done to facilitate their management and to guarantee equitable access to high-quality, safe and efficient health care for patients affected by conditions that require highly specialized care (Rodwell & Aymé 2014, 7).

In **Austria** care competences are shared between the federal level and the regional level. The system is characterised by "regionalized care provision within a regulatory framework determined at the federal level and delegation of statutory tasks to legally authorized stakeholders in civil society" (Hofmarcher & Quentin 2013, 40). The state and social health insurance funds contribute almost equal shares to the financing of the health care system (Hofmarcher & Quentin 2013, 17). Competencies are therefore also divided between regional entities of the social health insurances and across provinces (Trukeschitz et. al. 2012, 115). Health insurances play a significant role for the organisation of outpatient care delivery and in the primary care sector (Hofmarcher & Quentin 2013, 22). The fragmentation of responsibilities in health and social care makes it difficult to identify regional demands and to coordinate between the regions and different forms of care supply (outpatient and hospital care) (Hofmarcher 2013). The national structural plan sets the parameters for regional and local care provision. It is designed in cooperation with all important actors of the health system, including the insurance funds. Yet, the building of collaborations and partnerships between health and social care providers is not encouraged and is not yet a standard practice (Ladurner et. al 2011, 43). Although attempts are made to improve care provision for people with complex needs - i.e. chronically ill patients implementing structured disease management programmes remain a challenge in Austria (Hofmarcher & Quentin 2013, 22) and rare diseases are not covered by the programs.

The **Swedish** health care system is based on shared responsibilities. All three levels of Swedish government are involved in the health care system. The role of the central government is to establish principles of care provision and to set the political agenda for health and medical care. The mainly tax -

funded system is run by the county councils (Glenngård 2018). Municipalities are responsible for care of the elderly and disabled people. Social care is also mainly provided by the municipalities. Both, health and social care are supervised by the national Health and Social Care Inspectorate. Provisions are also covering the main caregivers of patients in need. Family member giving care to their relatives are paid at wages comparable to those of municipal employees. Caregiving is targeted at those in need of extensive care services, while others receive less and must rely on family members or pay for help privately (Palme et. al. 2003).

In **Romania** regional structures have been set up as well. The national level is responsible for the implementation of government health policy and the district (=judet) level responsible for ensuring service provision according to the rules set centrally (Vlădescu et. al. 2008, 16). Yet, critics point out, that decision-making in the Romanian health system is still in parts centralized, with administrative regulation and financial control concentrated at the national level (Vlădescu et. al. 2008, 22).

The most important planning tool in the health sector is the National Health Strategy (Vlădescu et. al. 2008, 23). The main objective in the area of service provision of the current strategy, which came into force in 2014, is to ensure equitable access to quality and cost-effective health services. Moreover Romania strives to strengthen planning capacities at all levels (national, regional, local), including the establishment of a planning unit within the Ministry of Health. The Strategy also mentions specific national plans targeted at distinct diseases. Rare diseases are among the areas addressed in the national health care plan (Vlădescu et. al. 2008, 24),

Compared to the other countries Romania has basic, but quite limited health and social care provision. Healthcare expenditures are covered by a comprehensive insurance package, including about 85% of the population who contribute to the health care scheme. Disabled people and children are eligible for insurance benefits due to their status. People working in agriculture, those not officially employed, self-employed or unemployed who are not registered for unemployment or social security benefits as well as Roma people who do not have identity cards are uninsured. They can only access minimum care packages (Vlădescu et. al. 2008, 570).

Because of financial shortcomings there is a shortage of service provision as well as health care professionals, affecting mainly rural areas (Blanco 2017, 20). Hence, international reports state, that Romania has "severe problems with the management of its entire public sector (Björnberg 2018, 19). It is recommended to improve "access as well as quality of care and its distribution between population groups and regional areas" (Blanco 2017, 18) and to set up a sustainable, more efficient funding scheme. In this context case management appears to be very promising, as it connects existing resources.

4.1.2. POLICIES ON RARE DISEASES

Recommendations on rare diseases launched by the European commission, such as the communication on rare diseases by the European commission in 2008, had an important impact on national policies. In many European countries these recommendations initiated the establishment of dedicated policy boards and structures in the field of rare diseases. Still it is important to see the differences in the European Member states, regarding the history of policy development and of the structures that have been set up in the field of rare diseases.

In Spain the initiative for the formulation of a National Strategy on rare diseases, following the recommendations of the European Commission, was launched by the Spanish Ministry of Health in the same year (Rodwell & Aymé 2014, 5). It sets out strategic aims the objectives and recommendations for the implementation of a national rare disease strategy in Spain. For the development of the strategy a multi-stakeholder committee was installed that included scientific societies, patient organisations, the Spanish ministry of health and representatives of the Health Departments of the Autonomous Communities. The Strategy for rare diseases as well as any other related measures or actions aimed at rare diseases are included in the Spanish National Health Budget. Main objectives and strategic aims addressed by the Spanish National Strategy on rare disease are: information on rare diseases, prevention and early detection, healthcare, therapies, integrated health and social care, research and education/training. Given the decentralised health administration of Spain the Strategy provides a framework and a set of recommendations to the regions that are in charge of its implementation.

Funds were allocated through a call for proposals opened to the Regional Governments in order to facilitate the implementation of the Strategy (Rodwell & Aymé 2014, 6).

As in the other European countries the designation of reference centres, eligible to join the ERNs, is an important task fulfilled by the national government. Centres of Expertise are not focused on rare disease solely but provide services for diseases that require special therapeutic techniques and services as well as rare diseases "which, because of their low prevalence, require a concentration of cases for their adequate care" (Rodwell & Aymé 2014, 7).

Part of the strategy on rare diseases was the creation of CREER in 2009, a centre for rare diseases in Burgos[5]. It is dedicated to a better coordination of rare disease expertise, research, innovation, professional training and the dissemination of information as well as awareness raising and support to other Spanish organisations. (Rodwell & Aymé 2014, 5) Moreover the centre has an important role for anticipating respite programmes for families, promoting the mutual knowledge and exchange of experiences between patients and families and providing information training concerning welfare.

In accordance with the national strategy some autonomous communities set up strategies of their own, as for example Extremadura (2010), Catalonia (2009), the País Vasco (2011) and Murcia (2018). Catalonia, one of the regions which we will focus on in depth, is aiming at the implementation of a regional master plan on integrated health and social care (Rodwell & Aymé 2014, 7).

Yet important structures for rare disease patients and regional action plans had already been developed before (Rodwell & Aymé 2014, 7).This is also due to the strong engagement of patient representatives in Spain. They built an infrastructure, offer guidelines and helplines for rare disease patients and run registries on services. Since 2000 they cooperate strongly with self-help groups for disabled people (Rodwell & Aymé 2014, 12).

[5] Centro de Referencia Estatal de Atención a Personas con Enfermedades Raras y sus Familias de Burgos (CREER) - "State Reference Centre for rare disease Patients and their Families")

In **Sweden** a national board responsible for the coordination of rare disease policies was set up in 2010. In 2011 the National Function for RDs (NFSD - Nationella Funktionen för Sällsynta Diagnoser) was founded. They promote the coherence and the coordination of health care resources for people with rare diseases, try to increase cooperation with the social insurance, employment services, social services, NGOs and other actors and contribute to the dissemination of knowledge and information and to the exchange of good practice and experiences. In contrast to the other countries there is no national action plan in place in Sweden. It was developed by different stakeholders and submitted to the government in 2012, but has still not been officially adopted. From the point of view of Rare Disease Sweden, there is an urgent need for a national plan of action for rare diseases to be put in place. This plan should have a comprehensive approach, bringing together and coordinating all the various activities for rare diseases centred on the needs of the patient (RD Action 2017b, 5).

Sweden employs a different definition of rare disease, pointing to the effects of rare disease, as severe lifelong disability. Therefore, Swedish patient organisations profit strongly from the collaboration with organisations for disabled people who have achieved the implementation of strong social rights for people with disabilities on a local level and allocation of funds on a national level. By the beginning of 2017 there will be a Centre for Rare Diseases on each of the university hospital sites in the country (RD Action 2017b, 1)

In Austria the initial starting point for rare disease policies did not come before the year 2011. At that time the "National Coordination Office for Rare Diseases" was established by the Austrian Ministry of Health. It is run by the Austrian Health GmbH (the national research and planning institute for public health care) and funded by the Austrian federal ministry of labour, social affairs, health and consumer protection. Funds are negotiated annually.

The centre aims at better care conditions for patients with rare diseases and at the support of coordination action and exchange between different stakeholders dealing with rare diseases. It was responsible for the formulation of the National Plan for rare diseases and its implementation. In this process two committees were installed in Austria: A multi-stakeholder group including experts and patient organizations and a strategic platform which includes delegates of the decision makers in the Austrian health care and social system.

Main aims addressed in the NAP are:

- The implementation of an adequate coding system for rare diseases, to allow for better epidemiology and reimbursement
- setting up encompassing registries on rare diseases
- the process of the designation of centres of Expertise in Austria (Rd Action 2017a, 2).

By 2018 two centres have been designated in Austria. Patient care in Austrian Centres of Expertise must be provided by a multiprofessional team comprising not only all the relevant, necessary medical personnel, but also non-medical personnel like social workers, etc. (RD Action 2017a, 3) in order to comply with the aim of holistic care. Moreover the provision of trainings on specific rare diseases is a prerequisite for the official designation as a Centre of Expertise in Austria (RD Action 2017a, 4).

Except for the services provided in Expertise Centres other specialised social services focusing on the needs of RD patients are not well developed in Austria. There is no official rare disease helpline operating in Austria, and there are currently no plans to establish one (RD Action 2017a, 6).

In Romania the National Committee for rare diseases (NCRD) was created in 2013. It is an interdisciplinary scientific body without legal personality. Yet, in contrast to Austria the National Action Plan in Romania was included in the National Public Health Strategy for 2014-2020 ("Prosperity of Health") (RD Action 2017c,1).The strategy initiated a regulatory, political framework that generates a system to integrate health and social services. Funding was originally approved in 2015 and 2016. Yet there is no budget dedicated to the implementation of the national plan which makes its implementation difficult (RD Action 2017c,1). By 2018 Romania started to implement a few issues of its national plan such as the national evaluation of Centres of Expertise (RD Action 2017c,2). By now six healthcare providers have been designated as centres of expertise.

There is a helpline in place dedicated to rare diseases in Romania. It is privately funded and is available to both patients and professional individuals. HelpLine is operated by the Romanian Prader Willi Association (RPWA) and it is part of EURORDIS' Network of Helplines (RD Action 2017c).

Comparing the development of policy instruments in the four countries we see, that Spain and Sweden appear to be ahead of Austria and Romania. Although both forerunner countries implement policies

very differently they both have a quite long tradition of policy development in the field of rare diseases. In Spain rare diseases was a broadly discussed topic very soon. In Sweden rare disease patients can build on the inclusive understanding inherent to the welfare tradition of the state, pushing for high living standards for disabled people and for equality and inclusion according to need. Therefore Sweden is providing generous support to vulnerable groups, comparably high funds and well established structures allow the inclusion of disabled people (as for example the Social service Law, individual care plans,…). That is why – even though Sweden is still lacking a National Action Plan on RD (Rd Action 2007b), the legal framework on social services and its funding is favourable for the inclusion of rare disease patients.

4.2. Opportunities and barriers for up-scaling

The successful implementation of integrated care models depends on how integrated care is designed to fit the local context and needs (Pavlickova 2018). To create "fitting" solutions challenges in the context of health and social care systems need to be identified and overcome and opportunities for up-scaling need to be developed. The following chapter summarises the findings of the expert interviews and (online) discussions, focusing on the main challenges that need to be addressed to develop up-scaling possibilities for integrated care solutions. Each level of intervention – the policy level, the organizational level and everyday life situations of patients and their family members – are analysed separately, bearing in mind that they influence each other.

As explained in Chapter 3 we take a comparative perspective, describing the general issues of integrated care that all countries selected for the up-scaling process share –albeit to a varied extent. Most of the findings are based on experiences reported for Austria, Spain and Romania. Only in some cases we refer to Sweden, whose stakeholders did not want to get more involved in the up-scaling process.

4.2.1. POLICY LEVEL

4.2.1.1. Fragmentation of political domains, federal states and regions

Rare diseases are a multidisciplinary field. Realising patient-centred care that takes into account the medical and the social needs of patients and their families requires the combined efforts of **policy makers from different policy domains**. Traditionally the organisation of medical care falls into the responsibility of the ministry of health whereas the organisation of social care lies with the Ministry of Social Affairs and Labour. In Austria and Spain these two domains are combined in one ministry. Yet, this fact does not automatically provoke a joint agenda in the fields of health and social care, as bureaucracy, management traditions and legal frameworks may still hinder a stronger cooperation across domains. The Ministries of Education are comparably rarely involved in joint boards on rare disease policies.

Ministries and the public administration are historically grown institutions that are governed by their specific logics. They have developed their specific perspective on societal issues and on how to solve them (often known as “silo thinking” in the social innovation context). This has two consequences. Firstly, the respective perspectives on societal issues, the legal frameworks and the funding schemes are not complementary. Because the political structures deciding on the underlying legal frameworks and the financial resources are not integrated, but can be rather conflicting or fragmentary a stronger coordination of political agendas and care organisation can be difficult. Secondly, because of the different approaches of the political bodies of different domains they have to find some common ground first.

In many European countries, involving the ones selected for the up-scaling process, steps for a stronger integration of the relevant policy fields have been taken. One example is the **national action plans on rare diseases** where experts from different ministries were involved. In Spain, this was a first step to establish a stronger cooperation between the ministries for social care, health and education. Yet, collaboration between the ministries is often only selective and in many cases still exceptional. Cross-policy initiatives and a strong involvement of policy makers from different ministries are rare. In order for cross-policy initiatives to become more mainstream policy makers need to get used to a new way of

decision-making, not primarily thinking of their own resort, but of the societal issues that needs to be solved, as experts stress (E12). Therefore experts concluded in the discussion rounds that it is still a long way to go from the design of cross-policy fields to the implementation of political bodies that effectively deal with them.

Besides, the interface between social services and benefits provided by **federal states, regions and the national level** often causes obstacles in delivering integrated care. From a political and administrative perspective the delivery of social and medical care is a highly complex endeavour that involves the national, federal and communal level. The strong involvement of regional or even communal policy makers and political/administrative institutions ensures that local needs will be met. Therefore, national policies need to allow local authorities, policy makers and providers of social and health care to address the issues that are most pressing on the local level. This in turn means that the local level needs to be involved as it has increased accountability for realising integrated care solutions. How the autonomy of regions and federal states can contribute to the implementation of innovative structures of service delivery can, for example, be seen in Spain where some regions have set up legal frameworks and services to provide integrated care. [6]

However, regionalism and federalism can also be an obstacle for the realisation of integrated, patient-centred care. Often legal frameworks and budgets are not set up in a way that enables collaboration across regional and communal borders. Thus, beneficial effects that could arise from cross-border collaboration are not realised. It is highly problematic when patients are not allowed to access the best specialised unit in their vicinity, because it is located within the administrative limits of another region or federal state. Inequalities between patients living in different areas and limitations in the accessibility of health and social care provision, depending on the region they live in, may also be caused by different funding schemes of health insurances and different legal frameworks. In Austria, for example, no nationwide uniform service catalogue for the reimbursement of medicines exists. Hence, the costs

[6] Detailed examples for Spain can be found in D 9.4 – Roadmaps for integrated care

people with rare conditions might have to handle are depending on their insurance and the place they live in (E5).

The difficulty of the collaboration and the coordination between the national and the local level can be illustrated by the example of the national strategy on rare diseases in Spain. This strategy has been implemented in 2008 and entails specific measures to improve the situation for rare disease patients. However, health and social care is in the responsibility of the autonomous regions and the national strategy only has the status of a recommendation but is not binding in any way. Despite that, the strategy is still one of the most important tools to improve collaboration between the regions and it is the only national strategy on health in Spain foreseeing financial transfers to the regions that are dedicated to the development of registries on rare diseases.

The **establishment of joint policy boards** in the field of rare diseases, following recommendations launched by the European commission was in many cases a great opportunity for policy makers, especially social and health ministries, to practice interdisciplinary policy approaches across domains. However, until now these boards do not often involve the ministry of Education. They are not only involving policy makers but also patient representatives, giving them a voice in designing policies on rare diseases and in this way, representing a possible way of advancing holistic patient-centred care models.

Another possible solution for better coordination across regional borders is to link existing **rare disease-related databases on regional, national and European level**. Besides patient-centred-databases, reporting on the number and prevalence of certain rare diseases and also allowing the exchange of information between service providers, databases on existing services are built. Yet, in many countries existing databases are not linked properly. Hence, databases also need to be harmonised and linked to each other. Databases are also an important tool for political planning since they can identify potential gaps in care infrastructures or care services. These databases render the problems of rare disease patients more visible and raise awareness for their needs (E1).

In recent years steps have been taken in most countries to improve existing regional databases, to establish national databases and to link them to European databases which will facilitate the exchange

of information on rare diseases. Here ERN's and Orphanet play an important role. The exchange of data will allow designing clear intervention plans on a national level (as well as on a regional level) as experts hope (E14) and promote collaboration.

Moreover, databases including information on the patients' treatments can be used to support continuity of care. Hence, especially in the field of rare diseases where the next centre of expertise might be just across the border a transregional collaboration among regions, but also among Member States is paramount to guarantee care that is well-accessible and of good quality.

4.2.1.2. Funding schemes and legal frameworks

The realisation of integrated, inclusive and patient-centred care is partly dependent on financial resources. One major obstacle for the funding of integrated care is that health care and social care have different funding schemes applied by different ministries, in most cases the Ministry of Health and the Ministry of Labour or Social Affairs. Even if the domains health and social policy are in the responsibility of one ministry, as currently in Spain and Austria, dedicated financial resources might be allocated differently. Hence, in all the countries in question the development of innovative and integrated care solutions calls for re-thinking traditional responsibilities and funding mechanisms. One important means in this context is a **coordinated budget for health and social affairs** that provides funding from one hand.

Also the division of responsibilities between federal states has implications for the accessibility of social services and financial transfers, which are often in the responsibility of the federal states. Dedicated budgets to set up integrated care approaches and, especially in Romania, **more budgets for social and health services**, possibly tied to collaboration, would increase the opportunities to realise integrated care and benefit the development of social and health care provision.

Furthermore, since social and health care regulations have emerged with a specific perspective on care, they are not complementary. Hence, there are gaps between social and health care that make it difficult for rare disease patients to obtain the care they need. In this context, a common approach towards regulation from social and health care policy makers is much needed. If a common approach is not

decided on providers with an integrated care approached, providing social and health care from one hand, might face difficulties fulfilling both regulations.

Heterogeneous funding schemes and legislations also affect the patient themselves. They might have to go through an intense process of evaluation to be eligible for certain social and health services and financial transfers. In the case of rare diseases it is highly problematic if a diagnosis is needed to assess certain services since not all rare conditions do have a specific diagnosis. And receiving one may take a long time Therefore, due to the specifics of the social or health care regulations not all patients with rare diseases are entitled to access all the services they would need. Policy makers point out that a **more informed evaluation of both the disease and its effects by social care professionals** who assess what kind of services would be needed, would be beneficial (E15). They need to know that disabilities in the case of chronic diseases do not get better, but could also get worse. Hence, policy makers working in the social field would highly benefit from a stronger exchange with health care professionals. At the moment they often lack important information on the diseases and their expected course. Predictable changes in the health situation of the patients are therefore not taken into account when assessing a patient's care needs. By providing information of the potential development of diseases they would be able to assess care needs better and to provide social services based on the expected course of the disease.

4.2.1.3. Change of policy makers

Despite the national actions plans on rare diseases most EU countries set up, rare diseases are not yet a mainstream topic within the political context. Therefore, the implementation of **innovative care solutions often depends on the will of key decision makers to support such endeavours**. In the countries involved in the up-scaling process, the awareness for rare diseases of policy makers has increased throughout the last years. Here the recommendations on rare disease policies launched by the European commission and the developing structure of European Reference Networks have had an important impact. However, this positive development is countered by the fact that political structures, binding documents and regulations addressing the issue of rare diseases and of integrated care are rare and the continuation of European activities in the field for now looks somewhat fragmented with the exception of the European resource Networks. For rare diseases this situation can be very problematic.

Whenever personally dedicated policy makers leave office, the issues of rare diseases are at risk of losing political support and thus dropping off the political agenda. To combat this dependency on the goodwill of key policy makers sustainable structures seed to be established. Binding legal documents promoting the issues of rare diseases and enabling and monitoring the realisation of integrated care are necessary.

To conclude the interviews as well as the focus groups with stakeholders have shown that all the stakeholders involved regarded coordination and collaboration on the political level as highly important to realise innovative care solutions and to facilitate ongoing implementation processes of integrated care. Nonetheless, this is also the area where they see that change will take immense efforts and is only achievable in the long-term since it involves re-thinking care and eventually over-coming power structures that have grown throughout the last decades.

4.2.2. ORGANISATIONAL LEVELS

The provision of health and social care for patients living with a rare condition faces a structural dilemma: Due to the low prevalence of their diseases rare disease patients have very specific care needs. Yet rare disease patients are a rather fragmented group. The care needs differ strongly among them. What they share is the experience of the difficulties how to deal with a rather fragmented care system (EURORDIS 2017). Therefore, a leading question that needs to be addressed in the up-scaling process is to what extent the needs of patients with rare diseases need to be incorporated into universal strategies to provide integrated and innovative care to those who need it (a kind of "**mainstreaming**") and on the other hand, if and how concepts of integrated and innovative care need to be **adapted and tailored to the particular needs** of this group of patients? (Gruber & Holtgrewe 2017, 2). This translates into the challenges of accessing (specialized) infrastructure, knowledge transfer and the coordination of care, the following chapter will deal with.

4.2.2.1. Accessibility of social and health services

The low prevalence of the disease implies that only a small number of (medical) care professionals specialize in researching and treating a certain rare disease. Therefore, establishing specialized units on specific rare diseases often goes hand in hand with the centralization of the (medical) care provision.

These specialised centres are connected via the **ERN-Network**. In the selected countries the process on the selection of ERNs differs strongly. In Austria, for example, experts on one hand criticise that the comparatively highly regulated and therefore slower process of designation of centres of expertise could cause Austria to have limited access to Europe-wide exchange of knowledge among the centres of expertise (E5). On the other hand, the high standardisation of the process should allow for a thoughtful selection of specialised services (E1) and hence guarantee high quality of treatment.

ERNs should allow for a better knowledge transfer within the European Union. Yet, the high specialisation in the field of rare diseases is a challenge for the **diffusion of knowledge on rare diseases to the local level of primary health care**. At the moment the mainstream social and health care system often lacks expertise on rare diseases. They lack the information on existing services and hence cannot advice their patients properly on which health centres are the best to provide them with information regarding pathology, treatments of their disease. This is an important problem to be solved, as doctors in the primary care sector are often the first contact point of patients and their families. Hence, for a first diagnoses and in order to avoid mistreatment, doctors need to have a basic knowledge on rare diseases and need to know where to find specialists. This is especially critical for undiagnosed patients who might have to wait long for a concrete diagnosis or do not receive the right treatment due to a lack of expertise of physicians. Mainstreaming of knowledge on rare diseases and of the knowledge on where to find the experts will not only support a better and earlier treatment of the undiagnosed patients but also builds ground for the establishment of specialized centres.

Some regional policy makers interviewed therefore point to the importance of building structures for the distribution of basic knowledge on rare diseases to professionals working on the local level and to pay special attention to the primary care sector (E3). The challenge they address is how to **better connect the primary care sector with hospitals and specialized services**. In some regions of Spain this is realized by setting up electronic platform for the training of professionals working in the primary care field. In Austria guidelines on rare diseases, which can be used by professionals in the primary care sector, exist. Yet, experts pointed out that they are far too complex (E3).

A very good example of improving the diffusion of knowledge on rare diseases on the local level is Spain's plan to implement a nation-wide network using the established ERNs as a starting point (E13). It

proposes to link reference centres already established with local and regional specialised health services. Thereby they should be integrated in complementary networks on different levels: One level of expertise, one level of health care specialists and another level for the primary consultation. By now not all regions are covered by services that are part of the ERN-network (E15).

While the close collaboration of highly specialized experts contributes to the quality of health care and the creation of knowledge and expertise on rare diseases, it poses a **challenge in terms of accessibility**. As Austrian experts interviewed reported, in particular smaller hospitals might have difficulties to provide care needed by patients with rare conditions and sometimes reject to care for them as they fear costly treatments they cannot afford (E2). Because of that patients living with a rare condition and their families might need to travel far to access specialised services. Thus accessing high quality health care can be more difficult for people with rare diseases than for patients with a common disease. This is, as the interviews showed, especially difficult in Romania, where small communities are often not well connected due to broken roads, and a lack of public transport. It requires money and time if patients have to travel (long) distances to see the respective professionals.

Another problem that needs to be addressed in the context of accessibility is how and where to **integrate social services**. Close vicinity of care infrastructure is especially important in the case of **services needed on the daily basis, including day care facilities and specialised schools.** While traveling to a centre of expertise does not pose as much of a problem, when it comes to medical appointments that are only required bi-annually, easy accessibility is of major importance when it comes to social services. Specialized speech therapists, social workers or day care centres and schools need to be in close vicinity of the place people are living in. Therefore, care structures on the regional level are of great importance for the provision of integrated care for rare disease patients (E13). Policy makers report, that the coordination between services and the integration of social services can be fostered by guidelines on commissioning social services. Here a stronger awareness of policy makers and bureaucrats for the importance of collaboration between health and social services, for multidisciplinary approaches, is very important.

Another problem is that **existing specialised school and social services might have too little capacities (E10)**. In some cases only private bodies offer adequate infrastructure, posing a considerable financial

burden on the parents. Moreover, patient representatives in Romania and Spain reported that if parents feel that the care situation at school cannot be organised according to their children's needs, children might not attend school at all and are deprived of education. In order to enable a better inclusion of children with complex conditions in the school system some regions in Spain set up **education guidelines (E8)**. Also screening questionnaires, assessing the needs of RD patients, can be used for in-schooling to facilitate social inclusion of children. Sweden is a positive example regarding the inclusion of children in the schooling system since patients and their families have the right to social support (including early childcare infrastructures) by their communities. Even though especially mothers also in Sweden tend to stay at home longer, if they care for children with a rare condition than mothers in Sweden do in generally (E27), they find adequate day care infrastructure easier than in Romania, Spain or Austria. Here, schools and kindergartens that fit the needs of children with rare diseases are often far away – especially in rural areas - and therefore not accessible on a daily basis.

4.2.2.2. Doctors-centred health care systems

The work of care giving professionals such as doctors, nurses or social care specialists is governed by specific professional logics and a hierarchically structured work organisation. The way these professionals are educated and trained affects how they see problems, how they address them, how they work with other care professionals and how they communicate with patients.

Especially the field of medical care in many countries is highly specialised and follows strict hierarchies. Nurses and social care providers are given fewer responsibilities than doctors. This poses obstacles to the realisation of holistic, patient-centred care. Doctors-centred health care systems have difficulties in realising:

- A multidisciplinary approach, including the expertise of different professionals in the medical field and the expertise of social care givers.
- a preventative approach, as doctors are traditionally trained to focus on the question how to cure illnesses, instead of taking a preventative approach.
- A patient-centred approach, trusting in the patients abilities to organise care according to their needs.

As chronic diseases cannot be cured health stabilisation and prevention as well as the reduction of the severity of symptoms are crucial for people living with a rare condition (E3). This calls for a stronger involvement of professional expertise from other health-related fields as well as from the social field, who can bring in new perspectives. This would not only have a positive effect on the care situation, but also take a burden away from doctors. As doctors also have administrative tasks to do, they often have or dedicate little time to explain the illness and its effects to the patients and their relatives (E25).

The experts interviewed report, that because of the high specialisation of health care, professionals often lack confidence in the patients' abilities to decide on their care needs. This problem can be named as a form of medical paternalism. Yet, a lack of competences to decide on their own care needs does not only arise from medical paternalism, but is an effect of the organisation of care. Often professionals do not have the time to listen to their patients and to explain the illness and its effects to the patients and their relatives. This affects the ability of patients and care giving relatives to take their own decisions.

Patient-centred care, as an alternative to doctor-centred care systems, would mean on one hand that professionals take patients' views into account when planning health care and that they acknowledge patients and their carers as experts on their condition who are able to express their needs and are capable of organising their own care situation accordingly. On the other hand patient-centred care can only be realised if patients are well-informed, obtain guidance and help from the experts involved and have an active voice in the organization of care. In this regard care settings that do not allow taking time for the patients and care professionals who do not conceive the patients as experts of their own life are obstacles for patient empowerment.

Moreover, especially patients with rare disease would benefit strongly from a **better exchange across disciplines** as they depend on the expertise of many different professionals. Yet, because of doctor-centred systems and specialization the realization of a multidisciplinary approach is challenging. First, specialization in the medical field calls for cooperation between doctors from different disciplines. Second, collaborations become even more challenging if specialists from the social field are involved as the highest importance and most recognition is usually attributed to the medical profession. Therefore the main power and responsibility lies with the medical profession which makes it difficult for non-

medical experts to be recognised. Social care professionals have a different perspective on the way diseases can be treated and on its effects in everyday live. Thus, the low recognition of this expertise by medical professionals poses a potential obstacle to the realisation of a more holistic vision of care.

One possible solution is to **provide social workers and nurses stronger competences** as it is done in the context of multi-professional teams (E3), which are installed by some health services and hospitals. They can foster the transfer of knowledge across medical disciplines and (if included) across social and medical disciplines. This is also a way to foster the recognition of other professional expertise in the organisation of care, which is an important element of holistic care. As a case manager interviewed mentioned, the first thing doctors have to lean is how to collaborate (E11). They have to recognise that they cannot be "experts for everything" but that they need to become expert's cooperative work instead. This is calls for a "cultural transformation" of care organization and (E12). In the literature this challenge is referred to as the problem of finding common language between professionals of different fields and expertise and a common approach to the integrated care (Kodner & Spreeuwenberg 2002).

4.2.2.3. Coordination of care and knowledge transfer across disciplines

To improve the coordination of care giving services and care giving professionals two challenges have to be met. The first one is knowledge transfer. This includes, as has been stressed above the recognition of expertise from different fields. The other one is the task of coordination itself.

Regarding the **exchange of knowledge across disciplines**, technical solutions – as platforms and databases – are important. Moreover guidelines are used for a better transfer of knowledge across disciplines. As policy makers in Spain report especially a more informed evaluation of the illness and its effects by social care professionals deciding on the social services people with rare conditions might need would be helpful. At the moment these institutions often lack important information on the diseases and their expected course. Predictable changes in the health condition of the patients are therefore not taken into account when assessing the patients' care needs.

Efforts also need to be invested in a **better coordination between** health and social **care services** and a coordination of different public health services within countries and regions. Of special concern in the field of rare conditions is the organization of **transition from childhood to adulthood**. Often health and

social care services are limited to a certain age. The better collaboration between these care providers can improve continuity of care, relieve patients from some burdens of organizing care and of explaining themselves and their situation in different care situations.

In the context of INNOVCare, in Romania, but also in some Spanish regions these problems have been addressed by offering case management for rare disease patients. Case managers in these regions have different roles and functions, depending on the intuitional framework where case management is implemented. Case managers organise the contact between specialists and within multiprofessional teams. They are a first contact point for patients and bring them in contact with (specialised) health and social services. Case management is considered by experts interviewed as a good working tool of integrated care, already established in some regions. Thus the further development of case management in the field of rare diseases is also recommended in the national action plans on rare diseases in Spain and in Romania. Experts point out, that for up-scaling case management it is necessary to have a clear concept of function and role of case managers. Moreover funding is still challenging.

4.2.3. PATIENTS AND THEIR FAMILIES

The opportunities for people with rare diseases or other complex needs to participate in social life and to organize everyday life are closely linked to their immediate social environment, their families. The social background, education and economic wealth of the family play an extremely important role in this context. Depending on their social environment and on the supportive services patients and their families find and can afford, they can develop capabilities of coping with their situation. Problems patients and their relatives need to cope with are, for example, time consuming care, difficulties in finding adequate support and psychological treatments, difficulties to stay in the labour market or to find adequate schooling facilities and a low recognition and little awareness for the needs of people with rare conditions. Without adequate help main care givers – mostly mothers - often stay at home to care for their children and have limited possibilities to follow regular employment (see also EURORDIS 2017).

Hence, an important element for the empowerment of patients and their families is their **active participation in social and economic life** as well as economic independency to the greatest possible

extent. However, due to physical and mental stress and little time resources of rare diseases patients, as well as of their care giving relatives, both have problems to keep a regular employment and to actively participate in social life.

Possible solutions to these problems are, as has been pointed out by the experts, general awareness raising and the empowerment of patients on one hand, and supportive infrastructure and counselling for care giving relatives, family members and professionals dealing with the needs of patients with complex needs (such as (kindergarten) teachers) on the other hand. The stronger integration into working life of care giving relatives and patients contributes significantly to the reduction of the risk of poverty, as experts point out and is therefore a very important step for empowerment and the realization of equal citizen rights and the active participation in social life.

The social environment of patients is manifold. It includes their families and friends, involving their main care givers, as well as school, day care facilities or employment. The latter are important **settings, structuring the everyday life of patients of different age**. Hence, awareness raising by providing information and support has to start in these settings.

For the realisation of an inclusive school system, **school infrastructures as well as conditions at school and curricula have** to be adapted to the needs of children with rare, chronic conditions. This includes time slots reserved for treatments the children need and assistance with daily care needs by the teachers. They need to have the possibility to follow medical and social therapies without missing school. Often these therapies are only offered to certain times, preventing the children from going to school. Despite inclusive school systems pedagogic staff in schools and kindergartens might know how to help children with their medication or might not be allowed to help them due to institutional regulations leaving parents with the need to care for their children alone.

Experts report a lack of awareness of and support for teachers, day-care teachers, work colleagues and school mates for the needs of people with rare conditions. Hence these professions would need support and encouragement in order to know how to deal with challenges of rare diseases. At the moment teachers, who are confronted with the needs of rare diseases pupils, often have to inform themselves about the effects of the disease. In this regard guidelines and standardised information for teachers on

the disease, as set up in some regions in Spain, could support the inclusion of pupils with rare conditions as they assure teachers of how to set up supportive structures and where to find help. Until now, both domains - health and education - often lack awareness for the importance that the interface between these two domains has for the implementation of inclusive care and health systems.

Empowerment is often considered a domain of patients themselves or of self-help-groups. Yet, patient representative's stress, that empowerment is a societal challenge. For the empowerment of patients, it is paramount that patients are not only given a voice to express their needs, but that they are also given a "space of safety", know that their concerns are of a general interest and that there are people who support them. Hence, raising awareness cannot only be task of self-help-groups, even if they have an important role in this field. In order to make the needs of patients and their families visible, the whole society needs to be involved. Therefore information needs to be spread to the lay public and especially to relevant decision makers. To improve the visibility of self-help groups is a stronger cooperation with patient organisations of disabled people. This has a strong tradition in Sweden and proven to have a positive impact on the implementation of supportive structures for patients with rare diseases.

Also for **family members** it is often hard to find support. On one side, they often face the challenge of taking the first step to ask for help, as patient representatives report. On the other side special offers - including training and information - for family members of patients with rare diseases are often limited and, as in Austria, offered by private organisations only. As caring has an impact on the physical and mental health of care givers (pushing wheelchairs, lifting patients ,..) it is crucial to focus more strongly on needs of family carers and to offer services and trainings which help them to stay healthy. Patients' representatives report that relatives would especially profit from an easier access to psychological help and from daily assistance to deal with their care duties and domestic tasks. Furthermore, it is of high importance to support family carers in developing a positive perspective and in seeing the potentials of their family member living with a rare condition. Without possibilities to reflect their situation it is more difficult to develop future perspectives that do not merely focus on the burdens of the disease of their relative. Especially in families in which a child is living with a rare disease his or her siblings need to be informed about the disease and offered psychological help to better cope with the family situation. Peer to peer coaching and regular meetings with other families living with similar situations will empower

patients and their families, providing them with a setting to talk and to relax. Peer to peer training addressing these challenges are offered by Ågrenska in Sweden and by CREER, in Spain. In Austria and Romania these services are offered by NGOs.

4.3. Scaleable Domains

Upscaling is a highly complex process. Still, many models of upscaling in the social innovation context imply a rather mechanical process of implementing good practices tested in one context in another. However, this simple transfer can only rarely be realised in real life since institutions governing the respective field are already in place and local actors might have started to develop ideas on their own. In the context of developing innovative care solutions for people living with a rare or complex condition we therefore identified three scaleable domains. We consider scaleable domains to be areas in which further steps need to be taken in order to realise more holistic, patient-centred and inclusive care for people living with rare conditions and beyond. The three scaleable domains are organised according to the levels that are crucial for up-scaling integrated care solutions, as discussed in the previous chapter. Thus, the three scalable domains focus on the implementation of a more connected logic of care delivery, overcoming disciplinary and regional boundaries.

1. On the **policy level** a move from a bureaucratic logic to a more holistic logic, allowing for a **coordination and cooperation of different policy levels** and different domains can develop synergies and build ground for a more holistic organisation of care.
2. On the **level of care delivery** professional and organisational divides need to be bridged so that stronger integration of health and social care into a networked logic can be realised. In order to improve the coordination of care between service providers the INNOVCare project has piloted possible ways to **professionalise the coordination of health and social services**.
3. For patients and their family members a more connected logic in the organisation of care helps them to be aware of their rights and fosters their capabilities of communication with care organisations and care professionals. This would allow patients and their main care givers to move from a sort of fragile logic of coping to a more **empowered logic of active participation**, to be more involved and capable of organising care themselves and to participate actively in social life.

4.3.1. COORDINATION AND COOPERATION ON THE POLITICAL LEVEL IN SOCIAL AND HEALTH CARE

To foster coordination and collaboration on the policy level a joint agenda of policy makers across disciplines and regional boundaries is needed. This will allow them to find a common language and a common approach to the issues they are facing (West & Poulton 1997).

Collaboration can be fostered by joint policy boards, which have been set up for the development of national action plans on rare diseases in many European counties. They can be seen as a first starting point for this agenda. Rare diseases in this sense are not just one target field but may be considered a piloting field for developing more connected health and social care policies and a way of policy making.

Learning how to collaborate can be facilitated by these experiences. Hence in order to establish stronger collaborations between policy makers it is fruitful to build on past experiences of successful collaboration. This can also be fostered by dedicated **trainings**, including patients, patients' representatives and policy makers. Such trainings and conferences, which support the exchange between patients and policy makers, will support the exchange across disciplines and between patients and policy makers, making the needs of patients with rare diseases visible. The involvement of patient organisations and patients is an important element when setting up integrated care structures in order to recognise their needs adequately. Moreover databases, including **information** on care needs and existing services, support evidence-based planning of service supply. Yet, sustainable and durable **structures** supporting joint political actions in the field of rare diseases still need to be developed further. This process is not easy and requires **personnel and financial resources**.

A dedicated budget to facilitate coordination between the relevant ministries and other important political bodies would be needed. It will increase the likelihood of the coordination and collaboration to be successful as funding does not have to be negotiated. Moreover, a coordinated budget for health, social care provision and education beyond co-funded pilots would allow a better exchange across disciplines and a more integrated perspective, and integrated resources do not have to be taken from one department. Pre-financing integrated care will also raise awareness for the need of integrated services amongst policy makers, as experts stressed. Another important measure for a stronger

collaboration on the political level is to have qualified staff in all the public institutions involved that is responsible for the communication and mediation between the organisations.

A **stronger involvement and accountability** of local administrations responsible for the organisation of care delivery is very relevant to foster the implementation of integrated care models. Up-scaling is most successful if it relates top-down and bottom-up approaches, developing a joint national agenda and recommendations on one hand and reaching out for a stronger involvement and accountability of local administration as well as patient organisations. Regional governments can, for example, facilitate the cooperation of health and social care services, for example, by making collaboration between those two domains a prerequisite in public procurement (as an example of a top-down-approach). On the other hand, integrated services, such as NoRo, Ågrenska and others, can foster awareness for the need of integrated care and improve the coordination patient organisations, regional and national policy makers.

To summarise, a better coordination and cooperation on the political level will not only benefit people living with a rare disease and their families, but can improve care provision for the general population. The integration of health and social care is an important step towards advancing the primary care sector and assuring the accessibility of care. Moreover, increased efforts that are put into political planning, knowledge and data-exchange between the policy fields of health and social care can improve quality and efficiency of health and social care services. The implementation of innovative care solutions such as case management and community nurses can improve the accessibility of primary health and social care services for regions with a low coverage.

4.3.2. PROFESSIONALIZATION OF THE COORDINATION OF HEALTH AND SOCIAL SERVICES

Professionalization of the coordination of care can be approached in different ways. Multidisciplinary teams, coordination protocols, educational protocols, integrated trainings and case management are some tools that may improve knowledge transfer and the coordination of care. Multidisciplinary teams foster a holistic view and allow creating services tailored to needs of RD patients (health, social, housing, transport, economic situation).

When it comes to the delivery of care interdisciplinary team work alone is no guarantee for improving patient outcomes (McCallin 2001). Constant support and input from management as well as an encompassing **training** in team skills are paramount to unfold the potential of multidisciplinary health and social care teams (ibid).

In inter-professional trainings, involving patients and care givers, professionals can learn how to collaborate in a multidisciplinary and interactive sense. These sorts of trainings are already provided by resource centres, such as Ågrenska, and can support the further development of integrated care networks. They support the building of multiprofessional teams and of a more integrated shared understanding of care in general. These trainings need to involve social and health care providers, as well as teachers and family members. In some cases even policy makers were involved in the trainings, learning about the needs of patients. These networks will raise awareness for the needs of patients with rare dieses and support knowledge transfer across disciplines, which is an important precondition of a more holistic and inclusive care system.

What is more, **mainstreaming of knowledge** on rare dieses will support the building of a network. It can be made accessible to care providers through understandable guidelines. Those guidelines have to entail a list of specialised experts and information on how to get in touch with them. Coordination protocols, including health, social and educational affairs would help professionals in these fields to assess the situation of RD patients properly and to know where to find further information, if needed.

In the frame of INNOVCare an integrated, patient-centred approach, offering case-management by one-stop shop resource centres for integrated care, was piloted and tested. As the interviews and literature research have shown, case management, specialising in **connecting** the social and health care services and patients can be implemented in very different ways. In the highly complex field of care organisation for people living with rare or complex conditions, case managers can emerge as new profession. They can act as a first contact point for patients and relatives, informing on the impact of the disease and about possible services. This can also answer to the problem that professionals in

primary care often do not know how to deal with rare diseases and therefore by default send patients to the hospital which may not always be necessary and may add to the overburdening of hospital services[7]. Furthermore, case managers help to coordinate professional teams as well as services to ensure continuity of care. This helps, together with a protocol of treatment to ensure continuity of care and a holistic view on care.

Case management services can be offered by resource centres and patients' organisations, as it was tested in the frame of INNOVCare. They can be installed in hospitals, and take the role of an intermediary between professionals, connecting and managing multiprofessional teams, as it is done in Barcelona. Case management can be provided by municipalities, connecting different services and helping patients and their families navigating the system and bureaucracies. Also, other forms of regional case management in regional hospitals or health centres or by community nurses can be considered. In the aggregated views of our experts, case managers do not necessarily have to have a certain profession although the medical professions tend to see upskilled nurses as best suited for that role as they put the focus on the health care related tasks of case managers. Some duties of case managers – such as scheduling a time table for treatments – can even be provided or at least supported by technical solutions and online-tools. In some Swedish regions, apps, that support RD patients the coordination of appointments, are being tested.

What is important, when setting up case management, is to think about the function case managers should have within the care system and therefore in which structures case management should be embedded. Here it is important to build on existing resources and find synergies with other health care innovations. An intervention plan which clarifies the competences and responsibilities of different professionals, including the role of the case manager, can help in this context.

[7] https://innovcare.eu/wp-content/uploads/2018/09/4.-Session-1-Economic-evaluation-of-INNOVCares-pilot-of-case-management-Peter-Lindgren.pdf

Catalonia's Care Model for Rare Diseases (Spain), for example, foresees the implementation of an integrated care network, connecting regional case management and hospital case management. First, hospital case management is to facilitate the exchange between multiprofessional teams, especially during diagnosis and hospital treatments. They organise care in the specialised field of medical care, including psychological treatments. Additionally, territorial case management is to be installed. Territorial case managers connect the sector of primary health care, social care services in the region and hospital case management and help to organise follow-up-treatments. Also, in Murcia first steps have been taken to set up an integrated care network, given a legal framework and concept by the Plan for Integral Attention To Rare Diseases in the Region of Murcia. It has a budget of 12 million euros spread over 3 years (2018-2020). It focuses on Nurse Case Managers which will be installed in each of the nine health areas of the region of Murcia, and work in close contact with the regional reference hospital.

Case management is generally considered a very demanding job as case managers are the ones who are responsible to “put the spokes in the wheel” of care provision, in order to make it turn smoothly. Hence case managers need tools to deal with this responsibility and the patients’ needs. Training for case managers, including supervision and peer-to peer-training and a well-defined role will help.

Moreover, case managers are only as good as their network is. Hence, case managers need an **infrastructure** including information systems and databases informing on services for rare disease patients, legal frameworks and a professional network.

In conclusion, the professionalisation of the coordination of care through case management or other innovative forms clearly benefits patients with a rare disease and complex needs and not least, their family carers. First, professionalised services for the coordination of care help to orient the patients and to inform them about their illnesses and about services they can access. Second, they reduce the time burden of doctors since they are providing patients with a first contact point. Third, they reduce the time burden for the coordination of care for patients and their main care givers. Fourth, professionalized services for the coordination of care improve the continuity of care and therefore contribute to better health outcomes. Lastly, the coordination of care is an important aspect of holistic care models focusing not only on medical care needs but also on the social needs of patients.

4.3.3. EMPOWERMENT OF PATIENTS, THEIR FAMILIES AND PATIENT ORGANIZATIONS

The most important issue for empowering patients and their families is the implementation of measures to ensure equal participation in social life. Participating actively in social life improves quality of life and supports the preservation of health. Inclusive schooling facilities and support measures that aim at better participation in working life to enable patients to live a self-reliant life are extremely important in this context. Experts in this context also pointed to the position of siblings, who are in need for support.

This means, at first, to develop a common understanding of equality and of inclusion. To ensure equal participation of people with varied opportunities and resources means to treat them specifically, according to their needs. Hence, timely and preventive support for patients and family members, including legal frameworks supporting inclusion in social and economic life, counselling, information, psychological treatments and social services need to be set up. Approaching a more inclusive society is a joint effort and cannot only be the task of patient organisations. Public efforts and inclusive services are needed as well as peer-to peer learning infra-structures for patients, their family members and professionals working with rare diseases patients, allowing them to build a **social network**. Patients' representatives interviewed therefore stress, that general awareness raising is needed the most in order to improve the situation of patients with rare diseases is. **Information** campaigns targeting traditional and social media as well as the lay public can create awareness and help spread information on rare diseases, who are in themselves a very heterogeneous group of patients. General awareness raising and lobbying will help to keep or to put the specific problems of people with rare diseases on the political agenda. For a better inclusion of RD patients and their families self-help groups and patient organisations which are most active in this field, need to have strong public support. For a stronger voice of patient organisations, a joint agenda of disability and rare diseases patient organisations can be beneficial, as the case of Sweden shows.

An important aspect of a self-reliant life is the ability of patients to decide how they want to organise care. Therefore, they need sufficient information on the diagnosis, the disease and the expected course of their disease. Potential tools to foster the ability of patients and their families to decide on their care needs are trainings for themselves and doctors. **Communication training** for physicians could improve

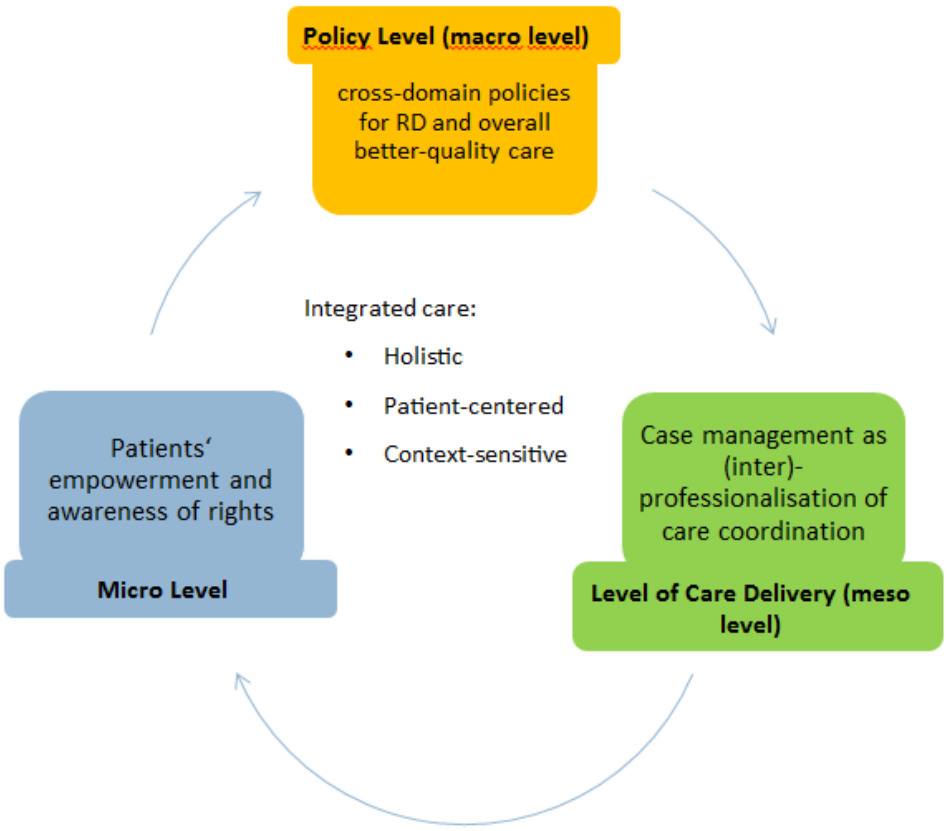
communication between them and their patients, giving patients a stronger voice in organising their care. In this regard case management plays an important role since case managers are a first contact point for patients living with a rare disease. They can facilitate the organisation of everyday life, provide information and put RD patients in contact with specialists. This first information and ongoing access to advice and counselling enables them to better manage their care situation. Moreover, better information of patients renders communication between physicians and patients easier as soon as legacies of medical paternalism and "learned helplessness" are overcome.

5. Summary: European perspectives on integrated care

The up-scaling report in hand is intended as a tool for policymakers and practitioners to decide on possible pathways to implement measures that facilitate integrated, holistic and patient-centred care. It describes obstacles and barriers to integrated care that need to be overcome as well as enablers that help approaching integrated, holistic and patient-centred care. Moreover, it identifies three scaleable domains representing areas in which further steps to realise more holistic, patient-centred and inclusive care settings for people living with rare conditions and complex needs can be taken.

The up-scaling report proposes a policy approach for up-scaling that combines bottom-up and top-down approaches and integrates stakeholders of different domains. This will ensure that all relevant intervention levels that structure the provision and delivery of integrated care are taken into account: The policy level, the organisational level and the micro level of the everyday life of patients with rare disease and their families.

FIGURE 3: INTERVENTION LEVELS AND ACTIONS



All three intervention levels benefit from a turn towards a more integrated and holistic logic. This much can be said across all kinds of local and regional contexts. On the policy level a more coordinated cross-domain policy approach can contribute to the realisation of integrated care, on the organisational level multidisciplinary approaches in care delivery as well as the professionalisation or care coordination through case management will be of great importance. On the micro level of the everyday life of patients integrated and inclusive care can be strengthened by the empowerment of patients and by raising awareness for their rights as patients. Overall, intensifying those efforts on all three intervention level will foster the inclusion of people with complex needs, increase their quality of life and potentially improve the overall quality of (health and social) care in the respective countries.

The INNOVCare project has contributed to improving the care situation for people living with a rare disease by developing and testing a case management approach offered by one-stop-shops. Case management facilitates the exchange between experts and the exchange of experts with patients and their families. In this sense it is a tool that can support networking within level and beyond levels. As the scaling report has shown, case management is only one possible tool to achieve a more coordinated, holistic approach. Besides case management training and formation, the spread of information on rare diseases, networking, coordination and collaboration involving stakeholders of all three levels will contribute to a more holistic logic of care delivery.

The up-scaling research done in the frame of the INNOVCare project and the pilot implemented in Romania provided a platform for exchange among stakeholders of different levels on a national as well as in a European context. In this sense the co-creation approach we decided on also supported the formation of a more interdisciplinary and holistic view of care on which the stakeholders involved can build on in the future[8].

As we have argued the field of rare diseases can be considered as a „liminal case“ to develop and test health and social services of better quality, to support more collaborative policies as well as multidisciplinary care work that integrates health and social care and thus realising a more patient - centred care approach. Policy plans on rare diseases have already led to collaborations across different policy fields at cross-political planning. They allow for a political integration of patient organisations. Moreover European pilots such as the INNOVCare project contribute to the distribution of multidisciplinary expertise across contexts. This turn towards an integrated logic of care delivery and organisation will not only benefit patients with rare diseases and complex conditions but potentially leading to an overall better-quality care.

[8] For an overview of the results of the INNOVCare project see here: https://innovcare.eu/bridging-the-gaps-between-health-and-social-care-results-of-the-eu-funded-project-innovcare-2018/

Based on the INNOVCare project's outcomes, the project partners have developed recommendations to implement mechanisms that ensure integrated care and integrated service delivery, coordinated between health, social and community services. In the recommendations it is made clear that only by implementing measures that foster integrated care the EU and European countries can achieve the principles set by the relevant policy frameworks of the European Union. These are the universal rights, such as the Universal Declaration of Human Rights, The United Nations Convention on the Rights of Persons with Disabilities and the European Pillar of Social Rights and expert recommendations aiming specifically at a better care delivery for people with rare diseases, such as the Commission Expert Group on Rare Diseases Recommendations to Support the Incorporation of Rare Diseases Into Social Services and Policies.

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Online resources:

http://www.imf.org

http://www.Rd-action.eu/rare-disease-policies-in-europe/

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