Improving organisation to improve care: ERN ReCONNET organisational reference model for systemic sclerosis patients' care pathway

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Abstract

Objective: To optimise the organisation of care and encourage the adoption of good clinical practices, the RarERN Path© methodology was designed within ERN ReCONNET. The aim of our work was to report the application of RarERN Path© on systemic sclerosis within the ERN ReCONNET centres, providing a feasible and flexible organisational reference model for optimising the systemic sclerosis care pathway in different countries.

Methods: RarERN Path© is a six-phase methodology which enables the creation of a reference organisational model co-designed on the basis of the expertise of different stakeholders. It foresees six phases, ranging from the map of existing patients' care pathways and patients' stories, to the consensus on a common organisational patient care pathways, and its key performance indicators definition.

Results: The agreed reference model highlights the importance of having an organisational flow for referrals that foresees how patients may access directly the specialised unit from the different referrals. Specific specialised visits were considered as mandatory to be organised and they included cardiologist, pneumologist, gastroenterologist, psychologist, nephrologist, dermatologist, wound care specialist/nurses and other healthcare professionals (such as nurses, social workers and nutritional counselling). Moreover, specific services related to therapy were highlighted as strongly recommended to be organised, mainly represented by infusion therapy and wound care, as well as occupation therapy and physiotherapy.

Conclusion: The organisational model emerged from our investigation emphasises that the organisation of specific services for systemic sclerosis treatment should be organised as a solid support for implementing the existing recommendations on systemic sclerosis management in real life.

Keywords

European Reference Networks, systemic sclerosis, patients' care pathways, organisation of care, organisational reference model, RarERN Path©, European Commission

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Significance and innovations

- The designed organisational reference model for systemic sclerosis (SSc) may support and promote a better organisation of care in different healthcare settings. Indeed, it clearly shows that European Reference Networks (ERNs) and other institutions (e.g. health authorities, international and national networks) can show how to improve the access to specialised centres, and in particular to treatments of rare disease (RD) patients, thus contributing to the promotion of equity of care in RDs.

- One of the main added values of the approach used to design the reference organisational model is represented by the multimethodology (including narrative medicine) and multistakeholder co-design approach. This can help in identification of different needs and priorities related not only to the disease-specificity but also to the different healthcare system frameworks

Background

Systemic sclerosis is a rare, chronic connective tissue disease characterised by autoimmune features, microvascular derangement and tissue fibrosis of the skin and internal organs (lungs, heart and gastrointestinal system).[1-3] The clinical picture is mainly characterised by Raynaud's phenomenon (RP), skin thickness, oesophageal dysmotility, digital ulcerations, interstitial lung disease and pulmonary hypertension.[4-6] The disease significantly impacts patients' personal (disfiguration) and functional (dyspnea, fatigue, weakness) aspects. Moreover, frequent follow-up visits and highly specialised care may be necessary to maintain disease control.[7-9]

In SSc patients, the disease heterogeneity and clinical complexity create a high level of stress, anxiety and uncertainty concerning their future. Several times, the optimal management of SSc is hampered by diagnostic delays and the lack of patient referrals to reference centres for appropriate diagnosis and effective treatment.[9] Thus, the complexity of SSc features requires a wide clinical approach, which is well represented in the updated European League Against Rheumatism/European Scleroderma Trial and Research (EULAR/EUSTAR) recommendations.[10]

The ERN ReCONNET – the European Reference Network on rare and complex connective tissue and musculoskeletal diseases (rCTDs) – [11] is one of the 24 European Reference Networks (ERNs)[12] established by the European Commission to improve the management and knowledge of rare and complex systemic connective tissue diseases, including SSc.

The main ERN mission is to improve the care of rare and complex conditions by sharing knowledge and promoting equity of care. One key area is the development of appropriate (clear and well-defined) patient care pathways (PCPs) to improve the care and management of patients with rare and complex diseases.[13]

The European Pathway Association defines the PCP as a 'complex intervention for the mutual decision making and organisation of care processes for a well-defined group of patients during a well-defined period'.[14] The PCP represents a valid tool to support both the application of clinical practice guidelines (CPGs) and the improvement of organisation of care, which is crucial to improve the care provided to patients. This issue is also highlighted during health emergencies, when patients and their families experience considerable limitations in access to care,[15] with consequences on their lives and clinical outcomes. Therefore, the organisation of patient care is pivotal to ensure an efficacious provision of services and support, and the lack of an efficient organisation can negatively impact the long-life journey of the disease and its impact on patients, their families and caregivers. Similar to other autoimmune diseases, the journey of SSc patients can become long and exhausting due to difficulties in accessing timely diagnosis, high-quality care and the overall management of such a complex disease throughout their lives.[16,17]

For this reason, a methodology was designed within ERN ReCONNET to optimise the organisation of care and encourage the adoption of good clinical practices. The methodology employed is RarERN Path©,[18] which provides a reference organisational model of the care pathway by bringing the expertise of different ERN centres and local patient advocates. Indeed, mapping SSc PCP organisations in different centres enables the collection, analysis and utilisation of both potential good practices and challenges in organisational structures. Definitively, designing an organisational model based on the CPGs can improve the application of the guidelines and deliver superior care.

The aim of our work was to apply for the first time the RarERN Path© approach to SSc within the ERN ReCONNET centres and to provide a feasible and flexible reference model for optimising the organisation of the SSc care pathway in different European countries.

Methodology

RarERN Path© is a six-phase methodology which enables the creation of a reference organisational model, co-designed based on the expertise of ERN centres, and the expertise and experiences of patients' representatives and individual patients. This method foresees six phases as follows (Figure 1):

Phase 1: Mapping of existing PCPs and patients' stories – aimed at getting the picture of the current practice in the organisation of care for a specific disease/disease area across the different ERN centres and at collecting patients' stories to capture how patients perceive the care they are provided as actors of a PCP, but also the impact of the disease on their lives.

Phase 2: Design of a common PCP – that foresees the elaboration and incorporation of the results of Phase 1 into a draft of a common PCP.

Phase 3: Consensus on a common PCP – in which different stakeholders discuss and reach a consensus on the final common reference organisational model for the specific disease PCP.

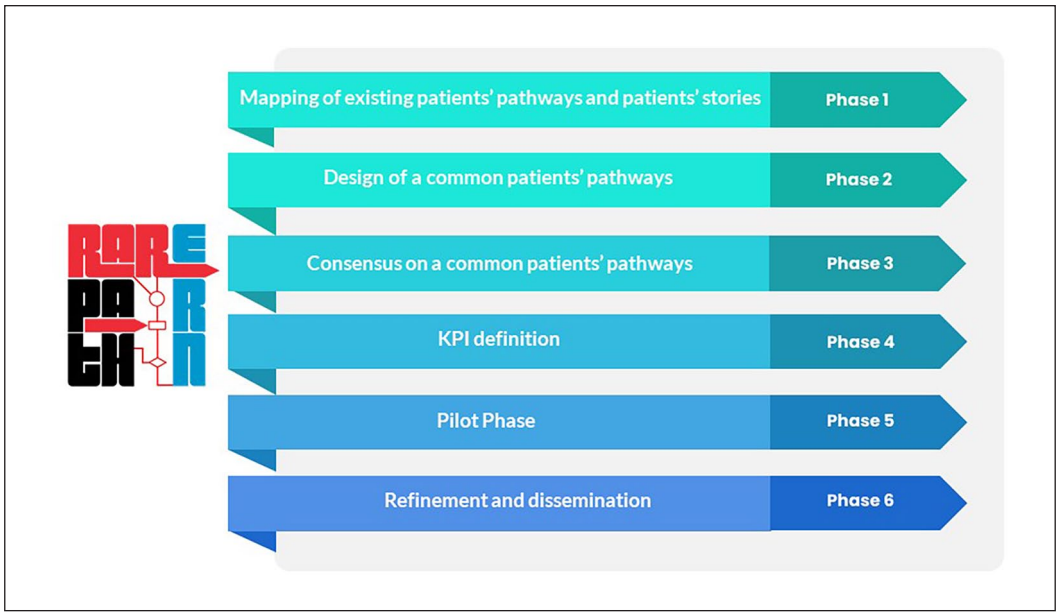

Figure 1. RarERN path phases.

Phase 4: Key Performance Indicators (KPIs) definition – in which specific KPIs are designed to assess the performance of the organisational reference model by different stakeholders to monitor the impact on the improvement of organisation in the healthcare providers that will adopt the reference model.

The method also includes Phases 5 and 6 that are related to a pilot phase and to a refinement of the organisational model based on the adjustments to be performed after the pilot phase. More specifically, during the pilot phase, the results from previous phases are applied in real life in a limited number of centres and the refinement process provides an adjustment of the model based on the feedback gathered from the pilot phase. As the results of Phases 5 and 6 are related to long-term data collection, they are not included in this article, which reports only the reference organisational model and the KPIs for SSc.

No Institutional Review Board (IRB) approval was required since the study did not involve patients' clinical data. Patients and clinicians have provided feedback and opinions as stakeholders involved in the SSc community. Moreover, in Phase 2, a narrative medicine approach was applied to anonymously collect the perspectives of patients regarding their point of view on the SSc PCP, which also related to their experience.

Target audience

The primary target audience is healthcare professionals who care for SSc patients, families and caregivers, as well as hospital managers and other stakeholders involved in organising care at local, national, European and international levels.

Application of RarERN Path© methodology in SSc

In Phase 1 (mapping of existing PCP and patients' stories), a specific questionnaire was sent to 20 ERN ReCONNET healthcare providers (HCPs). The questionnaire allowed collection of the current organisation of care for SSc, including organisational good practices and challenges. Each organisational care pathway was graphically reported into a flow chart illustrating the different phases followed (access, diagnosis, treatment and monitoring) in the single unit. The individual flow chart was reviewed and approved by each HCP representative to confirm that the graphical representation corresponded to the real-life organisation of the centre. In parallel, the narrative medicine approach was adopted to collect the views and perspectives of SSc patients from different countries, and this process was coordinated by experts in narrative medicine and patient engagement. The ePAG (European Advocacy Group) Advocate for SSc identified a panel of European SSc patients' representatives of the Federation of European Scleroderma Associations (FESCA) from Portugal, Spain, Belgium, Romania, Germany and Italy. The panel, together with the ePAG Advocate for SSc, co-designed a survey in English aimed at collecting patients' stories and translated the survey into different European languages to reach the widest number of SSc patients possible (Portuguese, Italian, German, Dutch, French, Romanian and Spanish). The survey was then launched across the SSc patients' community via email, social media and other RD community channels with the support of SSc patients' organisations. During Phase 2, the different flow charts of the ERN ReCONNET HCPs collected in Phase 1 were merged into a draft of a common PCP model. The draft included all the

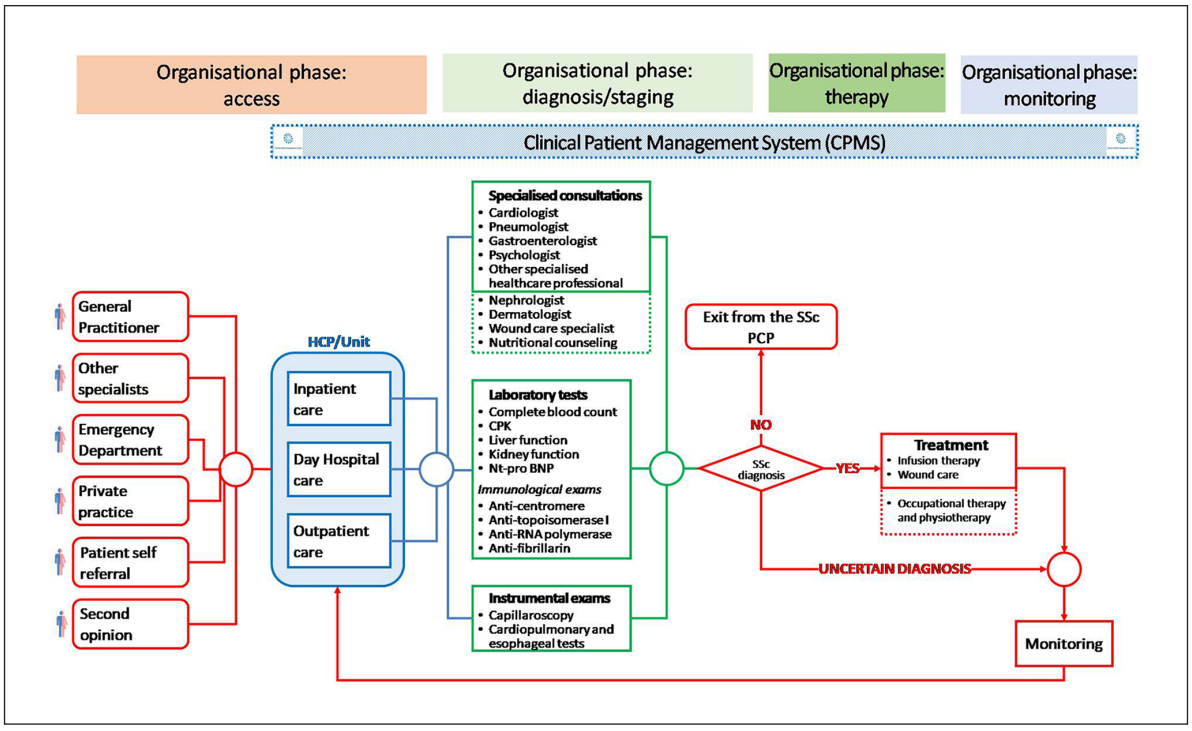

Figure 2. Organisational reference patients' care pathway model for SSc. Continuous lines indicate procedures considered as strongly suggested to be organised, while dashed lines indicate procedures not mandatory to be organised.

common elements and differences identified in the questionnaires, as well as the main challenges and suggestions related to the pathways that were raised by clinicians. The patients' stories ($n=203$) were elaborated by experts in narrative medicine, together with the panel of patients' representatives, to identify the common unmet needs as well as the main challenges and opportunities of the experiences lived by SSc patients (e.g. fear, anxiety, little awareness of being part of a specific pathway, long journey lived by patients before getting a diagnosis and the lack of adequate information for both patient and healthcare professionals). Besides the main topics identified in the stories, a quantitative analysis of the most common terms used in the stories was performed and integrated into the draft of the common PCP model.

The consensus on a common PCP foreseen in Phase 3 was implemented with the organisation of a dedicated interactive and multistakeholder workshop. After designing a common PCP and discussing the process together with the ERN ReCONNET Disease Coordinators for SSc (V.S., M.M.C. and I.G.), a multistakeholder consensus workshop took place in Pisa on 27–28 October 2019, during the 'Expert Panel Consensus meeting on Patients Pathways'. The workshop included participants from different stakeholder groups: patients' representatives, caregivers, researchers and clinician experts in SSc, hospital managers, experts in healthcare organisation and health economics, experts in patients' involvement and narrative medicine, experts in methodology and a policy officer of the European Commission. The primary focus of the workshop was to discuss the draft of the organisational reference PCP model resulting from Phase 2. More specifically, the discussion allowed all participants to analyse each individual phase of the PCP (access, diagnosis/staging, treatment and monitoring) from an organisational point of view, highlighting the main challenges, but also sharing the existing best organisational practices. Each phase of the model was explored to agree on the different services that should be organised to provide appropriate care to SSc patients, and the discussion was launched by addressing the question, 'Which of the services present in the organisational reference PCP model should be considered mandatory to be organised in healthcare providers taking care of SSc patients?' and by performing an agreement among the participants that were asked to indicate their agreement by voting ('yes' or 'no') for each PCP stage. The agreement on the mandatory organisational services to be organised for each PCP stage also included the related specific KPIs.

The design of the KPIs (Phase 4) was based on the results of the questionnaires and patient stories from the workshop discussion and by performing a final agreement with the ERN ReCONNET SSc Disease Coordinators.

Results

The SSc organisational reference model for PCP derived by the application of RarERN Path© methodology is represented in Figure 2.

The organisational reference model proposed by ERN ReCONNET intends to drive healthcare professionals and healthcare providers in adopting the 'optimised' organisational choices related to the provision of care to SSc patients. The results are reported following the main organisational phases illustrated in the organisational reference model.

Access to the centre

The main access points of SSc patients are related to different referrals that can include general practitioners, other specialists, emergency department, private practice, patient-self referral and also in case of a specific request of a second opinion. The agreed reference model highlights the importance of having an organisational flow for referrals (already diagnosed or with a suspicion of SSc) that foresees how patients may directly access the specialised unit from the different referrals. This organisation should include the allocation of specific flows, ensuring that all access points have a direct communication with the specialised unit. This includes the organisation of a fast-track access (e.g. in case of scleroderma renal crisis or severe new onset ulcer), to quickly refer the patient directly to the specialised unit, shortening the time to reach specialised care. Then, the triage may immediately identify patient's needs starting the best clinical and therapeutic procedure in different settings (i.e. inpatient or outpatient care, day hospital care, etc.).

Diagnosis/staging

For this phase, three main dimensions of the organisation of care were identified: specialist consultations, laboratory tests and instrumental exams. The mandatory specialised visits that were agreed for SSc include cardiologist, pneumologist, gastroenterologist, psychologist, nephrologist, dermatologist, wound care specialist/nurses and other healthcare professionals (such as nurses, social workers, nutritional counselling, etc.). A specific agenda of visits and blood and other investigations should be dedicated to SSc patients. For SSc, the following lab work was considered mandatory: complete blood count, creatine phosphokinase (CPK), liver function, kidney function, NT-proB-type Natriuretic Peptide (BNP) blood test, as well as anti-nuclear antibodies and specific antibodies (anti-centromere, anti-topoisomerase 1, anti RNA polymerase, and anti-fibrillarin antibodies, Anti-mitochondrial antibody). The mandatory instrumental examinations were capillaroscopy, chest computed tomography (CT) scan, lung function tests, cardiological and oesophageal tests. When the diagnosis of SSc is not confirmed, the patient will exit the SSc-specific organisational pathway and will be directed to a different pathway proportional to the alternative diagnosis.

Treatment

Specific services related to therapy must be organised: infusion therapy and wound care, as well as occupation therapy and physiotherapy.

Monitoring

When SSc is confirmed, or when the diagnosis is undefined and a tight follow-up is needed, the next clinical controls should be scheduled directly with the specialist, and the laboratory tests and instrumental exams as well with a planned schedule and timing of the follow-up. Moreover, centres adopting the reference model should request an international cross-border consultation via the Clinical Patient Management System (CPMS) to ERN ReCONNET for particularly complex and rare cases that benefit from a multidisciplinary and highly differentiated discussion. The organisational reference model was designed to ensure flexibility of application in the different healthcare settings, enabling the organisation to manage the various phases with a modular approach, considering the specificities and organisational constraints between different centres.

The putative KPIs identified can be grouped into two main categories: process and outcome indicators (summarised in Table 1). After application of the reference organisational model in the pilot phase, the KPIs will be updated and/or redefined.

Discussion

Our data show that an 'optimised' organisational reference model may be adopted for SSc PCP, suggesting that a rational and simplified approach to SSc patients' diagnosis and care is possible and may be shared among ERN centres.

Clearly, improving the organisation of care by means of structured PCP can bring numerous advantages, facilitating in clinical practice the application of CPGs provided by the main scientific societies.[19] The organisational structure emerging from the reference model suggests a clinical strategy which may be easily applied in practice and is in line with the existing clinical recommendations. The model provided by the application of RarERN Path highlighted that a rapid access to care is crucial for the management of a rare and severe disease like SSc. Moreover, the discussion of the PCP model emphasises that a structured and recognised organisational pathway for the access of SSc patients to the hospital specialised centre and to its structures (outpatient and/or inpatient care, day hospital care) is pivotal, as well as the organisation of a fast-track access to the dedicated unit. Notably, this organisational structure has already been deployed in Belgium, after adoption through multidisciplinary consensus meeting by the Flemish network on rare connective tissue diseases.[20] Adoption of this organisational structure in other European countries would be desirable to be followed.

Table 1. KPIs for monitoring the application of the reference model for SSc PCP.

Process indicators	Time to referral: average, median, min–max days of waiting time for accessing to the dedicated Unit in inpatient and/or outpatient
	Time to diagnosis: average, median, min–max days from first access to the SSc diagnosis
	Time to decision to treat: average, median, min–max days from first access to SSc therapy prescription
	Time to wait-listed: average, median, min–max days from prescription to infusion in different settings (hospitalisation or day hospital care)
	Number of SSc patients accessing the HCP (inpatient and/or outpatient and/or day hospital setting) per year
	Number of specialist consultations performed for SSc (cardiologist, pneumologist, gastroenterologist, psychologist) per year
	Number of laboratory tests performed for SSc per year
	Number of instrumental examinations performed for SSc per year
	Number of follow-up visits performed for SSc patient per year
Outcome indicators	Level of satisfaction of patients towards the organisation of care taking into account the reference organisational model for PCP
	Level of satisfaction of healthcare professionals towards the organisation of care taking into account the reference organisational model for PCP

Regarding the organisational phase of the diagnosis and disease staging, an agreement was reached on consultations by specialists from different fields (e.g. cardiologist, pulmonologist, gastroenterologist, desirably with experience in SSc) and exams that must be ordered (i.e. by means of a dedicated agenda) for SSc and those that are recommended to be scheduled. According to what emerged from patients' stories, the participants agreed to add the psychologist to the list of mandatory consultations. Regarding therapy, the focus was on organising infusion slots as well as guaranteeing treatment availability.

A prompt access to the centre should be always ensured to provide optimal and organised follow-up visits and exams for all SSc patients. For this reason, every centre should be organised to schedule the visits for monitoring the disease.

One of the main added values of the approach used to design the reference organisational model is represented by the multimethodology (including narrative medicine) and multistakeholder co-design approach. This can help in identification of different needs and priorities related not only to the disease-specificity but also to the different healthcare system frameworks.[21] The reference organisational PCP model is flexible and may be applied to different RDs. A practical example of the different settings in which the model can be applied is represented by the organisation of an outpatient wound clinic, which was discussed during the workshop. In fact, the outpatient wound clinic has been considered mandatory according to specific setting and resources available in the single centre. The service can be performed within the unit by a specialised nurse, or it can also be provided by an external unit (e.g. dermatologists). This example shows that the main scope of the organisational reference model is ensuring the provision of the best efficient service(s) to the patients, regardless of how it is implemented.

The organisational model emerging from our investigation emphasises that specific services for SSc treatment should be organised in line with existing recommendations on SSc management in real life.

Conclusion

The designed organisational reference model for SSc may support and promote a better organisation of care in different healthcare settings. Indeed, it clearly shows that ERNs and other institutions (e.g. health authorities, international and national networks) can lead the improvement of the access to specialised centres, facilitating the adequate treatment of RD patients, thus contributing to the promotion of equity of care in RDs.

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Author contributions

R.T., D.M., I.P., S.C. and G.T. conceived the paper and application of the RarERN Path methodology. V.S., M.M.C. and I.G. managed and supervised the work as ERN ReCONNET Disease Coordinators for SSc. All the co-authors contributed in the discussion of the reference model, as well as in revising the manuscript.

Authors' note

The Editor/Editorial Board Member of *JSRD* is an author of this paper; therefore, the peer-review process was managed by alternative members of the Board and the submitting Editor/Board member had no involvement in the decision-making process.

Disclaimer

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Ethical approval information, institution and number

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Patient and public involvement

ePAG and patients were involved in the work as members of working group for SSc PCPs.

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References

1. Benvenuti F, Zanatta E, Avouac J, et al. The power of the EUSTAR cohort: key findings to date and implications for management of systemic sclerosis patients. *Expert Rev Clin Immunol* 2020; 16(11): 1065–1074.
2. Varga J, Trojanowska M, Kuwana M, et al. Pathogenesis of systemic sclerosis: recent insights of molecular and cellular mechanisms and therapeutic opportunities. *J Scleroderma Relat Disord* 2017; 2(3): 137–152.
3. Volkmann ER, Andréasson K and Smith V. Systemic sclerosis. *Lancet* 2023; 401(10373): 304–318.
4. Cutolo M and Smith V. Detection of microvascular changes in systemic sclerosis and other rheumatic diseases. *Nat Rev Rheumatol* 2021; 17(11): 665–677.
5. Vanhaecke A, Debusschere C, Cutolo M, et al. Predictive value of laser speckle contrast analysis in systemic sclerosis. *Eur J Clin Invest* 2022; 52(1): e13672.
6. Di Battista M, Barsotti S, Orlandi M, et al. One year in review 2021: systemic sclerosis. *Clin Exp Rheumatol* 2021; 39 Suppl 131(4): 3–12.
7. Becker M, Graf N, Sauter R, et al. Predictors of disease worsening defined by progression of organ damage in diffuse systemic sclerosis: a European Scleroderma Trials and Research (EUSTAR) analysis. *Ann Rheum Dis* 2019; 78(9): 1242–1248.
8. Bellando-Randone S, Del Galdo F, Lepri G, et al. Progression of patients with Raynaud's phenomenon to systemic sclerosis: a five-year analysis of the European Scleroderma Trial and Research group multicentre, longitudinal registry study for very early diagnosis of systemic sclerosis (VEDOSS). *Lancet Rheumatol* 2021; 3(12): e834–e843.
9. Smith V, Scirè CA, Talarico R, et al. Systemic sclerosis: state of the art on clinical practice guidelines. *RMD Open* 2018; 4(Suppl. 1): e000782.
10. Kowal-Bielecka O, Fransen J, Avouac J, et al. Update of EULAR recommendations for the treatment of systemic sclerosis. *Ann Rheum Dis* 2017; 76(8): 1327–1339.
11. ERN ReCONNET, https://reconnet.ern-net.eu/
12. European Reference Networks, https://health.ec.europa.eu/european-reference-networks/overview_en
13. Statement of the ERN board of member stateson integration of the European Reference Networks to the healthcare systems of Member States, https://health.ec.europa.eu/system/files/2019-07/integration_healthcaresystems_en_0.pdf
14. About care pathways, http://e-p-a.org/care-pathways/
15. Talarico R, Aguilera S, Alexander T, et al. The impact of COVID-19 on rare and complex connective tissue diseases: the experience of ERN ReCONNET. *Nat Rev Rheumatol* 2021; 17(3): 177–184.
16. Saketkoo LA, Frech T, Varjú C, et al. A comprehensive framework for navigating patient care in systemic sclerosis: a global response to the need for improving the practice of diagnostic and preventive strategies in SSc. *Best Pract Res Clin Rheumatol* 2021; 35(3): 101707.
17. Farina N, Benanti G, De Luca G, et al. The role of the multidisciplinary health care team in the management of patients with systemic sclerosis. *J Multidiscip Healthc* 2022; 15: 815–824.
18. Talarico R, Cannizzo S, Lorenzoni V, et al. RarERN Path: a methodology towards the optimisation of patients' care pathways in rare and complex diseases developed within the European reference networks. *Orphanet J Rare Dis* 2020; 15(1): 347.
19. Talarico R, Marinello D, Cannizzo S, et al. An opportunity to harmonise the approach to patients' care pathways for rare and complex diseases: RarERN Path™. *Front Health Serv* 2022; 2: 935014.
20. Piette Y, Van den Bossche F, Aerts J, et al. Flemish network on rare connective tissue diseases (CTD): patient pathways in systemic sclerosis. *Acta Clin Belg* 2024; 79(1): 26–33.
21. Talarico R, Aguilera S, Alexander T, et al. The added value of a European Reference Network on rare and complex connective tissue and musculoskeletal diseases: insights after the first 5 years of the ERN ReCONNET. *Clin Exp Rheumatol* 2022; 40 Suppl 134(5): 3–11.