Original Manuscript

JSRD Journal of Scleroderma and Related Disorders

Journal of Scleroderma and Related Disorders
2021, Vol. 6(1) 96–101

DOI: 10.1177/2397198320953063
journals.sagepub.com/home/jso
SAGE

Requirements for systemic sclerosis expert centres in the Netherlands: A Delphi consensus study

Julia Spierings[1], Rita Schriemer[2], Sonja Dittmar[2], Lian de Pundert[3], Jeska de Vries-Bouwstra[4], Els van den Ende[5,6] and Madelon Vonk[6]; On behalf of the ARCH study group

Abstract

Introduction: Systemic sclerosis is a rare and complex disease. Optimal management of patients requires knowledge and experience and, importantly, intensive collaboration between hospitals and multidisciplinary teams. Definition and recognition of expert centres in systemic sclerosis is currently lacking, which complicates collaboration between centres and leaves patients poorly informed. The aim of this study was to develop a set of requirements for two types of systemic sclerosis centres in order to establish a nationwide structure for an optimal and transparent organization of care.

Methods: A three-round Delphi study was conducted among a panel of rheumatologists working at university or regional hospitals across the Netherlands. Prior to the final consensus round, a session with a patient panel (N = 22) was held. The results of this meeting were described in the last round for rheumatologists. Criteria were divided into five categories: (1) medical care, (2) case load, (3) collaboration, (4) research, (5) training of staff, and (6) other. In the first round, criteria derived from literature were proposed and participants could add criteria that were missing. For every item, participants could indicate if they thought the item should be included for two types of systemic sclerosis centres: (1) systemic sclerosis expert centre or (2) systemic sclerosis treatment centres. Consensus was reached when more than 85% of the panel agreed.

Results: In total, 47 rheumatologists participated in Delphi round 1, 35 in round 2 and 43 in round 3. Additional suggestions were added by the patient panel (n = 22). Consensus was reached for the requirements of systemic sclerosis expert centres (45 items) and systemic sclerosis treatment centres (29 items) including minimal caseloads of annual suspected systemic sclerosis cases and total patients in care.

Conclusion: Requirements of centres for systemic sclerosis care in the Netherlands were established in this study. Feasibility of certification should be evaluated next. Our proposed list can serve as a model for other countries.

Keywords

Systemic sclerosis, Delphi consensus study, quality of care, criteria, organizational structure

Date received: 10 June 2020; accepted: 5 August 2020

[1]Department of Rheumatology and Clinical Immunology, University Medical Centre Utrecht, Utrecht, The Netherlands
[2]NVLE, Dutch Patient Organization for Systemic Autoimmune Diseases, Utrecht, The Netherlands
[3]Department of Rheumatology, Haga Ziekenhuis, The Hague, The Netherlands
[4]Department of Rheumatology, Leiden University Medical Centre, Leiden, The Netherlands
[5]Department of Rheumatology, Sint Maartenskliniek, Nijmegen, The Netherlands
[6]Department of Rheumatology, Radboud University Medical Centre, Nijmegen, The Netherlands

Corresponding author:
Julia Spierings, Department of Rheumatology and Clinical Immunology, University Medical Centre Utrecht, Heidelberglaan 100, 3584 CX Utrecht, The Netherlands.
Email: J.Spierings@umcutrecht.nl

Introduction

Systemic sclerosis (SSc) is a rare, chronic and complex connective tissue disease.[1] Screening and treatment of disease-related organ and vascular complications and timely referral for advanced therapies can be challenging in this heterogeneous group of patients. Therefore, training of staff, interdisciplinary and multicentre collaboration and agreement on organization of health care services are paramount.[2] In order to build such an infrastructure, facilities and expertise available in centres need to be identified. Furthermore, insight in the level of expertise of centres is highly valued by patients.[3] Unfortunately, there is currently no uniform and transparent definition of expert centres for SSc. Consequently, collaboration between centres and multidisciplinary teams is complicated. Also, patients are not able to check whether they are treated in a centre that is engaged in managing this rare condition

In some other orphan diseases, requirements for expert centres have already been defined, are endorsed by scientific organizations and are applied in clinical practice. The Dutch organization of pulmonology and European Society of Cardiology, for instance, defined requirements for expert centres for interstitial lung disease[4] and pulmonary hypertension,[5] respectively. Also, care for patients with breast cancer, prostate cancer and rare types of cancer is already structured according to centres of expertise.[6–8] In 2014, the Dutch Society of Rheumatology published a guideline for SSc management and defined recommendations for referral to expert centres.[9] Yet, a widely accepted definition of SSc specialist centres is currently lacking. The aim of this study was to develop a set of requirements for SSc expert and treatment centres in order to establish a nationwide structure for an optimal and transparent organization of care.

Methods

Design

We used the Delphi technique in order to reach consensus about the requirements for expertise in two types of centres: SSc expert centres and SSc treatment centres. We defined a priori SSc expert centres as highly specialized centres provide a wide range of complex diagnostics and treatments and have deep knowledge about SSc. SSc treatment centres are hospitals that offer a selection of diagnostics and therapies and have dedicated multidisciplinary teams for SSc. SSc expert centres can fulfil the role of SSc treatment centres as well, but not vice versa.

Delphi technique involves a structured series of questionnaires (rounds) to gather information until consensus is attained.[10] The number of rounds was set on a maximum of three, based on previous Delphi studies. Between 6 January and 10 April 2020, rheumatologists participated in the online questionnaires. After the second round, we organized a live session with patients to explore their views on expert centres for SSc and to discuss the results of the two rounds. The patient panel meeting was held on 22 February at the University Medical Centre Utrecht. Informed consent was obtained from all participants.

Participants

A Delphi study was performed across rheumatologists working at both academic and regional hospitals in the Netherlands. We aimed to create a list of requirements that is supported by our Dutch colleagues; therefore, we invited all rheumatologists in the Netherlands to participate in the study. We approached them via the monthly newsletter of the Dutch Society of Rheumatology (NVR) and by email via the Dutch organization of systemic autoimmune diseases (SANL). Patients were approached on social media by the Dutch patient organization (NVLE). All patients wanting to participate could join the panel meeting.

Delphi rounds

In the first Delphi round, background information (sex, age, discipline) was collected. The questionnaire with requirements consisted of 40 items, ordered by categories: (1) medical care, (2) case load, (3) collaboration, (4) research, (5) training of staff and (6) other (see Supplementary Table 1). The items originated from criteria described in literature. This literature search was performed in PubMed and MEDLINE databases using the terms SSc or scleroderma combined with quality assurance, certification, requirements, outcome measures and care facilities. In addition, international and national SSc guidelines and expert definitions of other conditions were included. All English and Dutch papers were included and screened. Also, the set of requirements for expert centres for systemic autoimmune conditions according to the Dutch Federation of University Medical Centres (NFU), the Dutch society of regional specialist centres (STZ) and European Reference Networks (ERN) were used. One reviewer extracted items and discussed these with three other researchers.

To prevent wrong interpretation of the items, a description was provided for each item.

Participants could indicate if they thought that fulfilment of the items was required for SSc expert centres and for SSc treatment centres, using a Likert-type scale: (1) completely agree, (2) agree, (3) disagree, (4) completely disagree. Items about case load were open-text fields in the first round and four-item multiple choice questions in the second and third rounds. All items needed to be scored in order to complete the questionnaire.

In the first round, participants could suggest additional items in an open-text field, these items were added to the second Delphi round (see Supplementary

Table 1). Questionnaires were built in the online © Calibrum Surveylet software for online Delphi studies. For the last Delphi round, © SurveyMonkey software was used.

Consensus

Consensus was defined as agreement of at least 85% of the participants (completely agree and agree or disagree together with completely disagree). Items on which consensus was reached were removed; remaining items and additional items suggested by participants in the first round were included in the next round. In the second and third rounds, participants were informed about the response of the whole group. In the third round, results from the patient panel discussion were shown for each category.

Patient panel meeting

After two online questionnaires among rheumatologists, we organized a separate meeting with a patient panel. During this meeting, the patient perspectives on SSc expertise were discussed and items on which no consensus had been reached in the online rounds were addressed. The meeting started with a presentation of the background of the study, followed by the results of the second Delphi round. Patients were asked to vote on the items that were still subject of debate and could add new items. Results from the discussion and voting were summarized and added to the last Delphi round.

Data analyses

Characteristics and responses of rheumatologists were analysed using descriptive statistics.

Results

Participants

In total, 330 rheumatologists were invited to participate in the study, of which 14% (n = 47) completed the first round. Next, 35 completed the second and 43 the third Delphi round. Of all participants, 61% were female and 39% male, median age was 46 years (range: 34–63 years) and 89% of the participants were working as a rheumatologist, 11% worked as internist.

The patient panel meeting was attended by 22 patients, 86% was female, median age was 56 years (range: 29–80 years). The median disease duration was 4 years (range: 0.5–24 years), 50% (n = 11) of patients had the diffuse type of disease, 36% (n = 8) had limited cutaneous SSc and 14% (n = 3) did not know the disease subtype. Half of the patients were treated in an academic hospital, two (9%) in a regional hospital and four patients (18%) in both (shared care). Supplementary Table 2 shows the results of the discussion in the patient panel.

Requirements for SSc expertise

The panel of rheumatologists reached consensus for the requirements of SSc expert and treatment centres on, respectively, 45 and 29 items (see Table 1).

With regard to the SSc expert centres, the panel agreed that those centres had to be able to provide all therapies, including combination therapy for pulmonary hypertension and autologous stem cell transplantation, and collaborate with multiple disciplines and health professionals. SSc expert centres must have a team including a rheumatologist, pulmonologist, nephrologist, cardiologist, gastroenterologist, dermatologist, rehabilitation expert, psychologist, occupational therapist, specialized nurse, dietician, and a social worker engaged in SSc. These collaboratives with specialties do not necessarily need to be present within the centre, but can exist between centres. Multidisciplinary team meetings between specialists and centres involved should, however, take place regularly.

A structured multidisciplinary annual visit had to be offered at these centres. The minimal number of patients with suspected SSc should be 50 (62% of the participants) annually and >150 patients have to be in care (60%). The requirements regarding patient education, suggested by the patient panel, were adopted by the rheumatologist panel. Furthermore, SSc expert centres should have an expert status from the NFU. There was no agreement on the requirement of expert status according to the ERN (55% pro and 45% against) or STZ status (45% pro and 55% against).

With regard to SSc treatment centres, the panel agreed on 29 items. SSc treatment centres must have a team including a rheumatologist, pulmonologist, nephrologist, cardiologist, gastroenterologist, dermatologist, rehabilitation expert, psychologist, occupational therapist and a physiotherapist. Compared to the SSc expert centres, ability to provide highly specialized procedures, that is, right heart catheterization and combination therapy for pulmonary hypertension, is not required. Minimal caseload for suspected SSc was 10 annually and 45 for the total number of patients in care. In addition, an item about collaboration with general practitioners was adopted (see Table 1).

For both types of centres, requirements on training of staff and collaboration with other hospitals in the area and participation in national initiatives such as registries and the Arthritis and Research Collaboration Hub (ARCH) initiatives were included in the list.

Discussion

The aim of this study was to reach consensus among rheumatologist on requirements for expert centres for SSc in

Table 1. Consensus: requirements for SSc expert and treatment centres.

SSc expert centre		SSc treatment centre	
Medical care			
Diagnostics	*Agreement* (%)		*Agreement* (%)
Immunological tests	100.0	Immunological tests	97.9
Nailfold capillaroscopy	94.8	Nailfold capillaroscopy	95.8
Pulmonary function test	99.9	Pulmonary function test	97.5
Echocardiogram	99.5	Echocardiogram	85.7
HR-CT scan	100.0	HR-CT scan	90.4
mRSS	100.0	mRSS	90.5
MR heart	90.7		
Right heart catheterization	96.4		
Therapies			
Vasoactive medication	98.5	Vasoactive medication	93.5
Immunosuppressive therapy incl. CYC iv	100.0	Immunosuppressive therapy incl. CYC iv	92.8
Combination therapy for PH	99.9		
Autologous stem cell transplantation	85.5		
Facilities			
In-patient unit	100.0	In-patient unit	92.7
Intensive care unit	96.5	Intensive care unit	85.7
		Emergency medicine department	93.3
Case load (minimum)			
Annual number of suspected SSc cases >50	61.9	>10	70.3
Number of patients in care >150	59.5	>45	61.0
Collaboration			
Health care professionals			
Pulmonologist	99.9	Pulmonologist	97.9
Cardiologist	99.8	Cardiologist	95.8
Gastroenterologist	96.0	Gastroenterologist	97.6
Nephrologist	86.7	Nephrologist	86.8
Dermatologist	93.4	Dermatologist	88.0
Rehabilitation expert	96.0	Rehabilitation expert	88.1
Occupational therapist	88.3	Occupational therapist	87.7
Psychologist	87.5	Psychologist	85.3
Dietician	91.1		
Social worker	87.5		
Specialized nurse	99.9		
Multidisciplinary meeting	98.0	Multidisciplinary meeting	98.0
Annual visit to specialized nurse	100.0		
Structured multidisciplinary annual visit	88.1		
		Physiotherapist	85.1
Other			
Other centres	100.0	Other centres	92.0
Participation in ARCH and SANL	100.0	Participation in ARCH and SANL	90.4
Patient organizations	98.0	Patient organizations	89.4
		General practitioners	88.0
Research			
Participation in trials	97.5		
Initiation of trials	98.8		
Participation in registries	100.0	Participation in registries	100.0
Initiation of projects to improve quality of care	98.0		
Participation in international studies	88.8		
Training of staff			
Staff is trained in SSc every 2 years	93.4	Staff is trained in SSc every 2 years	87.1
Staff is trained in nailfold capillaroscopy	86.6	Staff is trained in nailfold capillaroscopy	90.5

(Continued)

Table 1. (Continued)

SSc expert centre		SSc treatment centre	
Staff is trained in mRSS	92.6	Staff is trained in mRSS	90.4
Experts provide training for other centres	96.0		
Other			
Centre fulfils requirements of NFU	85.4		
Patient education is integrated in usual care	100.0		
Patient education is available for any stage	100.0		
Patient education is provided by trained staff	91.4		

ARCH: Arthritis and Research Collaboration Hub; CYC: cyclophosphamide; mRSS: modified Rodnan Skin Score; NFU: The Netherlands Federation of University Medical Centres; PH: pulmonary hypertension; SANL: Stichting Auto-immuunziekten Nederland.

the Netherlands. Rheumatologists agreed on a set of requirements for two types of SSc centres with different levels of expertise. To our knowledge, this is the first study proposing such a list for SSc.

Establishment of consensus for two different types of SSc centres (SSc expert centre and SSc treatment centre) allows the design of a structure for medical services nationwide that fits the Dutch health care system. In this way, care for SSc patients with complex problems and/or poor prognosis in need of advanced diagnostic trajectories and intensive treatments can be concentrated in SSc expert centres. SSc care with low complexity can be done in SSc Treatment centres. Intensive collaboration between these centres is obviously essential, in order to align referrals and shared care. In this way, distribution of patients can be more balanced across the country, which will shorten travelling time to hospital appointments for many patients.

The agreed items for SSc centres are in line with set up requirements in other conditions. In pulmonary hypertension, the composition of multidisciplinary teams, case load and collaborative networks are defined as well in the set of requirements.[5,11] The set in our study also includes items about research, training and patient education, which are also adopted in the sets for prostate and breast cancer care.[7,12] Similar initiatives which are already implemented in practice in other conditions have shown to be feasible and well-accepted and have let to improvement of quality indicators in 220 centres after 5 years of follow-up.[13,14]

In order to put our work into practice, the list has yet to be acknowledged by the Dutch Society of Rheumatology, and all hospitals in the Netherlands could be evaluated according to our list. Creating a map with the level of SSc expertise in our country will increase transparency and enable patients and health care professionals to navigate the care system. Subsequently, certification of centres should be evaluated regularly.

Our study has some limitations. First, we made a selection of items we included in the questionnaires. This selection was, however, based on literature on existing requirements for centres for other conditions or categories of conditions (i.e. STZ requirements for systemic autoimmune conditions and recommendations for pulmonary hypertension expert centres). Furthermore, we invited participants to add suggestions for the item list in the first Delphi round. Also, we added items suggested by the patient panel. Second, representability could be questioned as the response rate after inviting all rheumatologists in the Netherlands was only 14%. Also, we were not able to record work setting of participants. We assume, however, that the study attracted both local and academically working rheumatologists with a special interest in the subject. Furthermore, the response (N=47) is acceptable for a Delphi consensus study, as the optimal sample for such studies is recommended to be between 10 and 50 highly engaged participants.[10] Another limitation of our study was that the case load acquired for both expert and treatment centres has a relative low consensus, which should be further debated before implementation takes place. Finally, we realize that different health care systems may use distinct approaches to organize care for complex and rare conditions like SSc, yet our national template can serve as an example for other countries as well.

In conclusion, our study resulted in a list of requirements that allows defining expert centres for SSc and identifying centres in the Netherlands that fulfil these requirements. In this way, patients and clinicians are better informed on where they can find the care they need.

Acknowledgements

The authors would like to acknowledge the Dutch Arthritis Foundation (ReumaNederland) for funding the ARCH initiative. Furthermore, they thank the Dutch patient organization (NVLE) for their efforts with regard to the patient panel session.

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.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship and/or publication of this article: This work was supported by the Arthritis Research and Collaboration Hub (ARCH) foundation.

ORCID iDs

Julia Spierings https://orcid.org/0000-0002-2546-312X

Madelon Vonk https://orcid.org/0000-0002-2266-9907

Supplemental material

Supplemental material for this article is available online.

References

1. Denton CP and Khanna D. Systemic sclerosis. *Lancet* 2017; 390(10103): 1685–1699.
2. Spierings J, van den Ende C, Schriemer R, et al. Optimal care for systemic sclerosis patients: recommendations from a patient-centered and multidisciplinary mixed-method study and working conference. *Clin Rheumatol* 2019; 38(4): 1007–1015.
3. Spierings J, Van Den Ende CHM, Schriemer RM, et al. How do patients with systemic sclerosis experience currently provided healthcare and how should we measure its quality? *Rheumatology* 2020; 59(6): 1226–1232.
4. NVALT bestuur. *Leidraad voor Kwaliteitsafspraken bij de introductie van nieuwe diagnostiek, medicatie of behandeling waarvoor Concentratie en Spreiding van Zorg wordt overwogen*. Dutch Association of Physicians in Chest Medicine and Tuberculosis, 2017, pp. 1–4.
5. Task Force for Diagnosis Treatment of Pulmonary Hypertension of European Society of Cardiology (ESC); European Respiratory Society (ERS); International Society of Heart Lung Transplantation (ISHLT) et al. Guidelines for the diagnosis and treatment of pulmonary hypertension. *Eur Respir J* 2009; 34(6): 1219–1263.
6. Wilson AR, Marotti L, Bianchi S, et al. The requirements of a specialist breast centre. *Eur J Cancer* 2013; 49(17): 3579–3587.
7. Valdagni R, Albers P, Bangma C, et al. The requirements of a specialist prostate cancer unit: a discussion paper from the European School of Oncology. *Eur J Cancer* 2011; 47(1): 1–7.
8. Casanueva FF, Barkan AL, Buchfelder M, et al. Criteria for the definition of Pituitary Tumor Centers of Excellence (PTCOE): a Pituitary Society Statement. *Pituitary* 2017; 20(5): 489–498.
9. Bonte-Mineur MECJ, Knaapen-Hans HKA, Meijs J, et al. Dutch guideline for treatment of systemic sclerosis. *Dutch Soc Rheumatol* 2014; (1): 1–59.
10. Hasson F, Keeney S and McKenna H. Research guidelines for the Delphi survey technique. *J Adv Nurs* 2000; 32(4): 1008–1015.
11. National Pulmonary Hypertension Centres of the UK and Ireland. Consensus statement on the management of pulmonary hypertension in clinical practice in the UK and Ireland. *Thorax* 2008; 63(Suppl. 2): ii1–ii41.
12. Biganzoli L, Cardoso F, Beishon M, et al. The requirements of a specialist breast centre. *Breast* 2020; 51: 65–84.
13. Brucker SY, Bamberg M, Jonat W, et al. Certification of breast centres in Germany: proof of concept for a prototypical example of quality assurance in multidisciplinary cancer care. *BMC Cancer* 2009; 9: 228.
14. Brucker SY, Schumacher C, Sohn C, et al. Benchmarking the quality of breast cancer care in a nationwide voluntary system: the first five-year results (2003-2007) from Germany as a proof of concept. *BMC Cancer* 2008; 8: 358.