Qualitative research in rheumatology: an overview of methods and contributions to practice and policy

Authors

Ayano Kelly, MBBS, FRACP [1,2,3,4]

Kathleen Tymms, MBBS, FRACP [1,3,4]

Kieran Fallon, FACSEP[1,4]

Daniel Sumpton, MBBS, FRACP [2,5,6]

Peter Tugwell, MD[7]

David Tunnicliffe, PhD[2,5]

Allison Tong, PhD[2,5]

Author affiliations

1. College of Health and Medicine, Australian National University, Canberra, ACT, Australia
2. Centre for Kidney Research, The Children's Hospital at Westmead, Sydney, NSW, Australia
3. Canberra Rheumatology, Canberra, ACT, Australia
4. Department of Rheumatology, The Canberra Hospital, Canberra, ACT, Australia
5. Sydney School of Public Health, The University of Sydney, Sydney, NSW, Australia
6. Department of Rheumatology, Concord Hospital, Sydney, NSW, Australia
7. Department of Medicine, University of Ottawa, Ottawa, Ontario, Canada

Corresponding author

Dr Ayano Kelly

Tel: +61415641166

Address: Centre for Kidney Research, The Children's Hospital at Westmead, Corner Hawkesbury Rd & Hainsworth St, Westmead NSW 2145, Australia

Email: Ayano.Kelly@anu.edu.au

Financial support

AT is supported by a National Health and Medical Research Council Fellowship (ID 1106716). The funders had no role in the study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Conflict of interest

All authors declare no relevant conflicts of interest.

Abstract

Patient-centred care is widely advocated in rheumatology. This involves collaboration among patients, caregivers and health professionals and is particularly important in chronic rheumatic conditions because the disease and treatment can impair patients' health and wellbeing. Qualitative research can systematically generate insights about people's experiences, beliefs and attitudes which patients may not always express in clinical settings. These insights can address complex and challenging areas in rheumatology such as treatment adherence and transition to adult healthcare services. Despite this, qualitative research comprises one per cent of studies published in top-tier rheumatology journals. A better understanding about the impact and role, methods and rigor of qualitative research is needed. This overview highlights recent contributions of qualitative research in rheumatology, summarizes common approaches and methods used, and outlines key principles to guide appraisal of qualitative studies.

Keywords

Qualitative research

Focus groups

Semi-structured interviews

Research design

Research methodology

Introduction

The need for patient-centred care is widely recognized in rheumatology, with shared decision-making being one of the cornerstone attributes in this paradigm (1-3). Clinical guidelines for rheumatic conditions have consistently emphasized that decision-making should explicitly consider the patients' values, preferences and needs (3-5). In addition, the World Health Organisation recommends that qualitative evidence is incorporated into the development of guidelines (6). This is particularly relevant because the interventions for patients with rheumatic conditions may have associated risks of complications and side-effects, and other impacts on the social, work-related and personal facets of their lives. Evidence on patients' experiences, attitudes and goals is thus needed to inform practice and policy.

Qualitative research methods can generate rich and detailed data to provide explanations and insights into the complexity of human behaviour and decision-making (7). Qualitative methods are used to generate hypotheses and address questions of 'how' and 'why'; whereas quantitative research methods are usually designed to test a hypothesis, and to answer questions of 'how much' or 'how often' (8, 9). Over the past decade, there appears to have been a growing number of publications of qualitative studies in biomedical journals across medical specialties, including rheumatology (10-13) (Table S1). However, qualitative studies remain a small percentage of published rheumatology research.

Between January 2015 to December 2019, qualitative studies comprised only 94 (1%) of the 8,484 original research articles published in the ten rheumatology journals with the highest impact factors in 2018 [Journal Citation Reports Social Science Edition (Clarivate Analytics,

2018)] (Annals of the Rheumatic Diseases, Arthritis & Rheumatology, Rheumatology, Seminars in Arthritis and Rheumatism, Therapeutic Advances in Musculoskeletal Disease, Osteoarthritis and Cartilage, Arthritis Care & Research, Arthritis Research & Therapy, Current Rheumatology Reports and Journal of Rheumatology. Only journals that publish original research articles have been listed.) (Table S1) In part, this may be because health professionals and researchers have little training and experience in conducting and appraising qualitative research methods or are uncertain as to how it can inform or impact upon practice and policy (14).

This overview will highlight recent contributions of qualitative research to care and policy in rheumatology and introduce qualitative research, including key approaches and appraisal of qualitative work.

Contributions of qualitative research to clinical practice and policy

In this section, we summarize the insights that qualitative studies have provided in clinically-relevant areas affecting multiple rheumatic conditions in adult and pediatric rheumatology: medication adherence,transition from pediatric to adult care and the experience and management of fatigue. We have also summarized additional, selected qualitative studies in selected rheumatic conditions (gout, rheumatoid arthritis, systemic lupus erythematosus) that are commonly managed by health professionals in rheumatology in Table 1.

Medication adherence

Non-adherence to medications is common across many rheumatic conditions with consequent impacts on patient morbidity and even mortality (15-18). Qualitative studies have been conducted in patients with rheumatic conditions to elucidate their experiences of and attitudes towards medications (19, 20). For example, patients with inflammatory arthritis are motivated to take disease modifying anti-rheumatic drugs in an attempt to return to their normal life and avoid future disability, though many often view their medications as a "necessary evil" with "toxic" side effects and uncertain efficacy (19, 21, 22). Medications may be perceived as a confronting reminder of their sickness and a threat to their health and well-being (19, 21, 23). For this reason, patients may decide not to take medications in order to regain control of their health and minimize lifestyle intrusions (23, 24). Patients can also be overwhelmed by the burden of deciphering multiple and sometimes conflicting information sources in order to make informed decisions about medications (19, 23, 25, 26). Studies have suggested that physicians can mitigate fears by facilitating shared decision-making and providing a supportive environment that allows them to voice their concerns about their medications (19, 26-28).

Transition from pediatric to adult healthcare

As young patients with rheumatic conditions transition from pediatric to adult care they must establish relationships with new clinicians, navigate different health care facilities and an adult model of care during a turbulent time of physical, social, vocational and psychological growth and change (29, 30). This is particularly relevant in rheumatology as many young people with juvenile-onset rheumatic diseases continue to have disease activity or significant sequelae in adult life (31). Qualitative studies in adolescents transitioning to adult rheumatology care highlight the challenges they face. They describe feeling abandoned and

ill-prepared to face a health care setting that is perceived to be sterile, de-personalized and uninviting (32-34). The transition process could be isolating if healthcare staff in the adult clinic focused only on medical aspects of care, with little consideration of psychosocial impacts of the condition (32, 35, 36). Patients can feel overwhelmed by the expectations to attend clinic appointment without their parents or to handover their own clinical information to new adult providers (34). In contrast, they felt more confident and secure when given an opportunity to become familiar with the adult physician and clinic, if information on the patient's knowledge and understanding of their disease was clarified, documented and handed over, and if they had the support of a specialist nurse in an adult clinic (30, 35). Patients undergoing transition to adult care appreciated a flexible approach that was tailored to their willingness and ability to take on more responsibilities and involvement in adult care (34, 35). Qualitative research has been used to help design and evaluate a transitional care program that incorporates the need for gradual and prepared transfer, regulated parental involvement and an adapted setting for adolescents (34, 37). Qualitative studies demonstrate the importance of transitional care programs to include familiarization, joint clinics, nursing support, adequate transfer of information and the provision of care that addresses the psychosocial priorities of young people.

Experience and management of fatigue

Fatigue is a common and debilitating symptom, and is of high priority to patients with rheumatoid arthritis (RA) (38). Previously it was rarely addressed in clinical practice as a treatment target in long-term care of patients with RA (39). However, in the last few years rheumatologists have become more aware of fatigue in the clinical setting for example with the increasing use of patient-reported outcomes that evaluate fatigue (40). A semi-structured

interview study with patients with RA found that fatigue permeated multiple aspects of life including work, leisure, family roles and led to feelings of uselessness and loss of self-esteem (41). The frustration, irritability and loss of control from fatigue negatively impacted on relationships. Fatigue was overwhelming, unpredictable and much more intense than the tiredness they felt prior to the onset of RA. Participants felt their fatigue was dismissed by health professionals, assumed that it was not treatable and that they had to manage it on their own. The findings from this study were used to develop a conceptual model of fatigue (42), a patient-reported outcome measure for fatigue (43), and a randomized controlled trial of cognitive behavioural approaches taught to nurses and occupational therapists in rheumatology care teams in order to improve fatigue (44). These studies highlighted the need for health professionals to address fatigue and ways to involve a multi-disciplinary team in supporting patients with this debilitating symptom.

Common approaches and methods used in qualitative health research

Qualitative research is inherently subjective as the purpose is to elicit opinions and understand human behaviour. The data are co-constructed between the researcher and participants. Theory may be used to inform the design and approach of the study. For some approaches, including grounded theory, the study is designed to generate theory from the findings. There are several specific approaches (i.e. strategies of inquiry, theoretical frameworks) that are used as a basis of conducting qualitative research (45, 46). These approaches can guide the procedures for participant selection, data collection and analysis. Although many approaches exist, five of the most common approaches in health research (47) are grounded theory (48), ethnography (49, 50), phenomenology (51-53), case studies (54), and narrative research (55). The differences in these approaches are summarized in

Figure 1 with illustrative examples of qualitative research in rheumatology (56-60). General characteristics of participant selection, data collection and data analysis that can apply to several qualitative approaches are described below. Researchers may design their study based on a single specific qualitative approach, or not specify a single approach but still use procedures that may be encountered within multiple approaches (45). Qualitative methods can also be used as part of mixed methods research and is discussed below.

Participant selection

Qualitative research typically involves an in-depth inquiry within a selected population. Purposive sampling is often used, which involves selecting participants who are relevant to the research question with the aim of including a diversity of relevant perspectives (8). Other sampling strategies include snowballing, which requires participants to identify other potential participants (61). This approach may be useful for including individuals who are hard to access, or with specific expertise or divergent opinions. These sampling methods are preferred to convenience sampling, which involves recruiting participants who are the most easily accessible (8) (e.g. consecutive patients from a single rheumatology clinic who are willing to participate in the study), because it is less likely to capture a broad range of perspectives.

Sample sizes are guided by the methodological approach, study design, participant population, research question and available resources. As a guide, semi-structured interview studies may have 30-60 participants depending on the amount of data obtained per interview (62). In grounded theory, 20-30 participants of each population included in the study is generally reported to be adequate. For phenomenological studies that require interviewing

each person multiple times, 6-10 participants may be sufficient (62). For focus groups 6 to 8 participants is recommended to optimize group interaction, with 3 to 5 groups for each characteristic of interest (e.g. based on a clinical diagnosis) (63). Rather than having pre-specified sample sizes, recruitment in qualitative research may cease when saturation is achieved (61). This is defined as when the collection and analysis of new data no longer elicits new insights.

Data collection

Semi-structured interviews and focus groups are commonly used in qualitative health research (64). These methods are effective in eliciting individual experiences, preferences and values to inform clinical practice. Focus groups capitalize on group interaction and allow participants to talk to each other, compare points of view, brainstorm ideas, and can be used to capture interpersonal language, culture and dynamics (8, 64). The facilitator keeps the group on task, asks probing questions, and encourages participation of all members of the focus group. Semi-structured interviews may be particularly useful when discussing sensitive issues or if participants are concerned about maintaining their confidentiality (64). The interview guide includes open-ended questions pertinent to the research questions (64). The guide is designed to encourage participants to openly pursue their perspectives in detail. Audio or visual recording and transcription of interviews and focus groups ensures that data is comprehensively captured for further analysis and can be complemented by field notes that capture contextual details, non-verbal communication and interactions within a group setting as well as initial reflections of the qualitative researcher (8).

Data may also be collected through observations or documents. Observations are a way of gathering data by systematically watching events and people to study their relationships and routine behaviours and is frequently applied in studies using ethnographic methodology (64). In qualitative health research this is particularly suited to understand how organizations work or how different members in the healthcare environment interact with each other (64). Observations may be covert or overt, and may involve the researcher as a participant or non-participant in the environment (64). Documents include printed and electronic material such as diaries, newspaper articles and organizational and institutional reports. These may be used for historical or policy studies or to evaluate health care organizations or programs (8, 65).

Data analysis

The analysis of qualitative data generally seeks to develop a comprehensive understanding and description of the phenomenon being investigated. The output of qualitative research differs depending on the methodology. For example, thematic analysis will yield themes (patterns of shared meaning that together give a comprehensive picture of the population of interest's experience) (66), ethnographic studies are designed to provide insights into the behaviours and perceptions of a sociocultural group (49), phenomenology seeks to describe a phenomenon from the lived experience of individuals (67) and grounded theory develops a theory arising from the data (48). The processes used in qualitative data analysis involve data reduction (by coding and identifying meaningful sections of the data into labels), data organization (in which codes are collected and sorted) and interpretation (where data are analysed to understand meaning, and codes are categorized, compared and emerging themes or theories are developed) (64). Data analysis should be an iterative process which involves

cycles of data collection, analysis and then resumption of data collection again to further explore and challenge emerging themes or theories (61).

Qualitative analysis software manages qualitative data and provides efficient methods for storing, organizing and retrieving qualitative data (8). These programs however, cannot conduct the analysis of the data. Investigators must create their own codes and interpret their data. The interpretive nature of qualitative research inevitably means that the researcher's background, knowledge and values can influence the analysis of the data. Several methods can be used to ensure the results accurately reflect the spectrum of the participants' perspective. These include member checking (sharing preliminary findings with participants to check whether their viewpoints are accurately captured), investigator triangulation (incorporating input from team members in the analysis, especially from different backgrounds) and reflexivity (reflecting on personal experiences and biases using a diary or field notes in relation to the data analysis) (8, 61).

Combining qualitative and quantitative research methods

A study may also use a mixed methods approach, which is a distinct research methodology where both qualitative and quantitative data are collected. Mixed methods research requires an integrated analysis and the use of rigorous qualitative and quantitative research methods (68). Mixed methods research can be classified into three core mixed methods designs: convergent (where qualitative and quantitative data are collected and analysed simultaneously within a single phase) (69-71); sequential explanatory (where quantitative data is collected first, then qualitative data collected to explain the quantitative findings) (72); and sequential exploratory (where qualitative data is collected, a feature such as a new

instrument or intervention is built, and then the feature is tested quantitatively) (43, 73, 74). These core mixed methods designs can be built into more complex research designs such as within a randomized controlled trial (41, 44, 75, 76) and is recommended for process evaluations of complex interventions (77). Before a trial, qualitative studies could generate hypotheses for examination, help develop and refine the intervention or outcome measures (80, 81) or enhance patient recruitment (82, 83). During a trial, qualitative methods could examine whether the intervention was delivered as intended, explore the participants' responses to the intervention and understand processes of implementation and change. After a trial, qualitative research can explain reasons for positive or negative findings of the trial, variations in effectiveness amongst trial participants, assess the acceptability of the intervention or be used to generate further questions or hypotheses (84).

Reporting and appraisal of qualitative research

The Enhancing Quality of Transparency of Health Research (EQUATOR) network recommends using the Consolidated Criteria for Reporting Qualitative Health Research (COREQ) as a guideline for reporting of qualitative research using interviews and focus groups (85). There are several other guides available for the conduct and evaluation of qualitative research (8, 61, 64, 85-87) including the American Psychological Association's Journal Article Reporting Standards for Qualitative Research (JARS-Qual) (88) which also provides guidance on how to structure a qualitative manuscript. However, the appraisal of qualitative research remains contentious, and there is debate as to how and even whether quality can be legitimately judged (64, 89, 90). There is no empirical evidence to indicate which criteria are critical and how to assess them (64, 90). The framework by Lincoln and Guba, addresses the rigor of qualitative research based on four criteria: credibility,

confirmability, dependability and transferability (91). The links between the COREQ reporting items and these constructs of rigor are shown in Table 2.

Credibility – Are the findings trustworthy?

Credibility refers to having confidence in the truth of the findings (analogous to internal validity in quantitative research) (87). Readers may be confident that the findings are a credible if the researcher provides a comprehensive and sensible explanation of the data. Comprehensiveness includes ensuring that the true breadth and depth of the phenomena in question were explored. Breadth of data can be captured using purposive sampling, continuing to sample until data saturation and the final sample size. Depth of data may be gauged by reviewing the question guide and duration of the interview or focus group to determine if they allow the participant to discuss the topic of interest in detail, and whether efforts were made to provide a setting that allowed participants to feel comfortable to express their opinions. In addition, triangulation in qualitative research allows a generation of deeper and richer insights. This includes using multiple data sources (data triangulation), data collection methods (data triangulation) or involving multiple researchers (investigator triangulation) in the analysis of data (8). Involving multiple researchers in coding can ensure that findings adequately capture all aspects of the data. A clear and insightful presentation of major and minor themes provides a final check on the comprehensiveness of data findings. Member checking allows participants to provide feedback on preliminary findings and ensures that findings are a sensible interpretation of their experiences.

Confirmability – Are the findings linked to the data?

Confirmability refers to the extent to which findings of the study are shaped by the data and are not a misinterpretation of findings by the researcher (analogous to objectivity in quantitative research) (87). This can be demonstrated by showing raw data such as quotations and linking them to findings from the study. The researcher may describe self-reflexivity whereby they recognize and reduce any undue influences on their interpretations of the data. Interpretations of data can also be confirmed using multiple data coders, triangulation and member checking, as noted in relation to credibility (87).

Dependability – Is the process auditable?

Dependability is analogous to reliability in quantitative research (87). Due to the nature of qualitative research it is not possible for another researcher to fully replicate a qualitative study. However, a rigorous and systematic approach to qualitative research can be followed with a coherent link formed between the findings and methods used in the study. Audio or video recordings, transcription of data and the use of qualitative software for coding allows transparent and auditable documentation of the research process (8). The raw data and analysis can thus be reviewed by others.

Transferability – Are the findings relevant to other contexts?

Transferability describes the degree to which themes or concepts from a qualitative study can be applicable to other contexts (analogous to external validity in quantitative research) (87). By providing details about participants' characteristics and study setting in enough detail (termed 'thick description'), readers of qualitative research can determine whether the findings may be applicable to their own setting. In addition, comparing the results of the

study with other studies in different populations or to existing theory can also help demonstrate the broader relevance of the study findings.

Conclusions

Qualitative studies have the potential to generate a deep understanding of people's experiences, motivations, beliefs, goals, expectations and needs. In rheumatology, evidence from qualitative studies has made a unique and valuable contribution to practice and policy. Qualitative research can be systematic, rigorous, and evaluated using the principles of credibility, confirmability, dependability and transferability. We suggest that further qualitative research is needed in rheumatology to address evidence gaps regarding patient priorities in the management of rare rheumatic conditions, co-ordination and integration of care amongst health care professionals and education about psychosocial impacts of disease. Incorporating insights from qualitative studies into clinical care, policies and trials can help promote patient-centred care to improve outcomes for patients with rheumatic conditions.

Tables and figures

Table 1. Examples of qualitative studies in rheumatology

Table 2. Appraisal of qualitative studies

Figure 1. Five key qualitative approaches used in health research

Supplementary material

Table S1. Qualitative research in the top ten rheumatology journals by impact factor

References

1. Barry MJ, Edgman-Levitan S. Shared decision making--pinnacle of patient-centered care. N Engl J Med 2012;366:780-1.

2. Voshaar MJ, Nota I, van de Laar MA, van den Bemt BJ. Patient-centred care in established rheumatoid arthritis. Best Pract Res Clin Rheumatol 2015;29:643-63.

3. Smolen JS, Landewe R, Bijlsma J, Burmester G, Chatzidionysiou K, Dougados M, et al. Eular recommendations for the management of rheumatoid arthritis with synthetic and biological disease-modifying antirheumatic drugs: 2016 update. Ann Rheum Dis 2017;76:960-77.

4. van der Heijde D, Ramiro S, Landewe R, Baraliakos X, Van den Bosch F, Sepriano A, et al. 2016 update of the asas-eular management recommendations for axial spondyloarthritis. Ann Rheum Dis 2017;76:978-91.

5. Ringold S, Angeles-Han ST, Beukelman T, Lovell D, Cuello CA, Becker ML, et al. 2019 american college of rheumatology/arthritis foundation guideline for the treatment of juvenile idiopathic arthritis: Therapeutic approaches for non-systemic polyarthritis, sacroiliitis, and enthesitis. Arthritis Care Res 2019;71:717-34.

6. Downe S, Finlayson KW, Lawrie TA, Lewin SA, Glenton C, Rosenbaum S, et al. Qualitative evidence synthesis (qes) for guidelines: Paper 1 - using qualitative evidence synthesis to inform guideline scope and develop qualitative findings statements. Health Res Policy Syst 2019;17:76.

7. Kuper A, Reeves S, Levinson W. An introduction to reading and appraising qualitative research. BMJ 2008;337:a288.

8. Giacomini MK, Cook DJ. Users' guides to the medical literature: Xxiii. Qualitative research in health care a. Are the results of the study valid? Evidence-based medicine working group. JAMA 2000;284:357-62.

9. Johnson SR, O'Brien KK. Qualitative methods in systemic sclerosis research. J Rheumatol 2016;43:1265-7.

10. Paley J, Lilford R. Qualitative methods: An alternative view. BMJ 2011;342:d424.

11. Saketkoo LA, Pauling JD. Qualitative methods to advance care, diagnosis, and therapy in rheumatic diseases. Rheum Dis Clin North Am 2018;44:267-84.

12. Borreani C, Miccinesi G, Brunelli C, Lina M. An increasing number of qualitative research papers in oncology and palliative care: Does it mean a thorough development of the methodology of research? Health Qual Life Outcomes 2004;2:7.

13. Tong A, Winkelmayer WC, Craig JC. Qualitative research in ckd: An overview of methods and applications. Am J Kidney Dis 2014;64:338-46.

14. Goguen J, Knight M, Tiberius R. Is it science? A study of the attitudes of medical trainees and physicians toward qualitative and quantitative research. Adv Health Sci Educ Theory Pract 2008;13:659-74.

15. Hsu CY, Lin YS, Cheng TT, Syu YJ, Lin MS, Lin HF, et al. Adherence to hydroxychloroquine improves long-term survival of patients with systemic lupus erythematosus. Rheumatology 2018;57:1743-51.

16. Jaleel A, Saag KG, Danila MI. Improving drug adherence in osteoporosis: An update on more recent studies. Ther Adv Musc Dis 2018;10:141-9.

17. Scheepers L, Burden AM, Arts ICW, Spaetgens B, Souverein P, de Vries F, et al. Medication adherence among gout patients initiated allopurinol: A retrospective cohort study in the clinical practice research datalink (cprd). Rheumatology 2018;57:1641-50.

18. Pasma A, Schenk CV, Timman R, Busschbach JJV, van den Bemt BJF, Molenaar E, et al. Non-adherence to disease-modifying antirheumatic drugs is associated with higher disease activity in early arthritis patients in the first year of the disease. Arthritis Res Ther 2015;17:281.

19. Kelly A, Tymms K, Tunnicliffe D, Sumpton D, Perera C, Fallon K, et al. Patients' attitudes and experiences of disease-modifying anti-rheumatic drugs in rheumatoid arthritis and spondyloarthritis; a qualitative synthesis. Arthritis Care Res 2018;70:525-32.

20. Rai SK, Choi HK, Choi SHJ, Townsend AF, Shojania K, De Vera MA. Key barriers to gout care: A systematic review and thematic synthesis of qualitative studies. Rheumatology 2018;57:1282-92.

21. Shaw Y, Metes ID, Michaud K, Donohue JM, Roberts MS, Levesque MC, et al. Rheumatoid arthritis patients' motivations for accepting or resisting disease-modifying antirheumatic drug treatment regimens. Arthritis Care Res 2018;70:533-41.

22. van Tuyl LH, Hewlett S, Sadlonova M, Davis B, Flurey C, Hoogland W, et al. The patient perspective on remission in rheumatoid arthritis: 'You've got limits, but you're back to being you again'. Ann Rheum Dis 2015;74:1004-10.

23. Nota I, Drossaert CH, Taal E, van de Laar MA. Patients' considerations in the decision-making process of initiating disease-modifying antirheumatic drugs. Arthritis Care Res 2015;67:956-64.

24. Salt E, Rowles GD, Reed DB. Patient's perception of quality patient--provider communication. Orthop Nurs 2012;31:169-76.

25. van Tuyl LH, Plass AM, Lems WF, Voskuyl AE, Kerstens PJ, Dijkmans BA, et al. Discordant perspectives of rheumatologists and patients on cobra combination therapy in rheumatoid arthritis. Rheumatology 2008;47:1571-6.

26. Shaw Y, Bradley M, Zhang C, Dominique A, Michaud K, McDonald D, et al. The development of resilience among rheumatoid arthritis patients: A qualitative study. Arthritis Care Res 2019.

27. Pasma A, van 't Spijker A, Luime JJ, Walter MJ, Busschbach JJ, Hazes JM. Facilitators and barriers to adherence in the initiation phase of disease-modifying antirheumatic drug (dmard) use in patients with arthritis who recently started their first dmard treatment. J Rheumatol 2015;42:379-85.

28. Salt E, Peden A. The complexity of the treatment: The decision-making process among women with rheumatoid arthritis. Qual Health Res 2011;21:214-22.

29. Tattersall R, McDonagh JE. Transition: A rheumatology perspective. Br J Hosp Med 2010;71:315-9.

30. Howland S, Fisher K. Looking through the patient lens - improving best practice for young people with juvenile idiopathic arthritis transitioning into adult care. Springerplus 2015;4:111.

31. Hersh A, von Scheven E, Yelin E. Adult outcomes of childhood-onset rheumatic diseases. Nat Rev Rheumatol 2011;7:290-5.

32. Ostlie IL, Dale O, Moller A. From childhood to adult life with juvenile idiopathic arthritis (jia): A pilot study. Disabil Rehabil 2007;29:445-52.

33. Wells C. Wellness in the midst of disease: A narrative analysis of growing up with rheumatic conditions [dissertation]. Minneapolis: University of Minnesota; 2015.

34. Hilderson D, Eyckmans L, Van der Elst K, Westhovens R, Wouters C, Moons P. Transfer from paediatric rheumatology to the adult rheumatology setting: Experiences and expectations of young adults with juvenile idiopathic arthritis. Clin Rheumatol 2013;32:575-83.

35. Shaw KL, Southwood TR, McDonagh JE. User perspectives of transitional care for adolescents with juvenile idiopathic arthritis. Rheumatology 2004;43:770-8.

36. Tunnicliffe DJ, Singh-Grewal D, Chaitow J, Mackie F, Manolios N, Lin MW, et al. Lupus means sacrifices: Perspectives of adolescents and young adults with systemic lupus erythematosus. Arthritis Care Res 2016;68:828-37.

37. Hilderson D, Moons P, Van der Elst K, Luyckx K, Wouters C, Westhovens R. The clinical impact of a brief transition programme for young people with juvenile idiopathic arthritis: Results of the don't retard project. Rheumatology 2016;55:133-42.

38. Wolfe F, Hawley DJ, Wilson K. The prevalence and meaning of fatigue in rheumatic disease. J Rheumatol 1996;23:1407-17.

39. Repping-Wuts H, van Riel P, van Achterberg T. Rheumatologists' knowledge, attitude and current management of fatigue in patients with rheumatoid arthritis (ra). Clin Rheumatol 2008;27:1549-55.

40. Fautrel B, Alten R, Kirkham B, de la Torre I, Durand F, Barry J, et al. Call for action: How to improve use of patient-reported outcomes to guide clinical decision making in rheumatoid arthritis. Rheumatol Int 2018;38:935-47.

41. Hewlett S, Cockshott Z, Byron M, Kitchen K, Tipler S, Pope D, et al. Patients' perceptions of fatigue in rheumatoid arthritis: Overwhelming, uncontrollable, ignored. Arthritis Rheum 2005;53:697-702.

42. Hewlett S, Chalder T, Choy E, Cramp F, Davis B, Dures E, et al. Fatigue in rheumatoid arthritis: Time for a conceptual model. Rheumatology 2011;50:1004-6.

43. Nicklin J, Cramp F, Kirwan J, Urban M, Hewlett S. Collaboration with patients in the design of patient-reported outcome measures: Capturing the experience of fatigue in rheumatoid arthritis. Arthritis Care Res 2010;62:1552-8.

44. Hewlett S, Almeida C, Ambler N, Blair PS, Choy EH, Dures E, et al. Reducing arthritis fatigue impact: Two-year randomised controlled trial of cognitive behavioural approaches by rheumatology teams (raft). Ann Rheum Dis 2019;78:465-72.

45. Tesch R. Types of qualitative analysis. In: Qualitative research: Analysis types and software. London: Routledge; 1990:77-92.: Routledge.

46. Creswell JW, Creswell JD. Qualitative methods. In: Salmon h, neve c, felts dc, marks a, editors. Research design: Qualitative, quantitative, and mixed methods approaches. Los angeles, sage publications; 2018:179-211.

47. Creswell J, Poth C. Qualitative inquiry and research design: Choosing among five approaches. 4th ed. Los angeles (ca): Sage publications; 2017.

48. Corbin J, Strauss A. Basics of qualitative research: Techniques and procedures for developing grounded theory. 3rd ed. Thousand Oaks CA: Sage Publications; 2008.

49. Reeves S, Kuper A, Hodges BD. Qualitative research methodologies: Ethnography. BMJ 2008;337:a1020.

50. Atkinson P, Coffey A, Delamont S, Lofland J, Lofland L. Handbook of ethnography. London, UK: Sage Publication; 2001.

51. Hammond M, Howarth J, Keat R. Understanding phenomenology. Oxford, UK: Blackwell Publishing; 1991.

52. Moustakas C. Phenomenological research methods. Virding a, editor. California, sage publications; 1994.

53. Van Manen M. Phenomenology of practice: Meaning-giving methods in phenomenological research and writing. Morse j, editor. Walnut creek: Left coust press inc; 2014.

54. Yin RK. Case study research : Design and methods. 5th edition. Los angeles: Sage publications; 2014.

55. Riessman CK. Narrative methods for the human sciences. Los angeles: Sage publications; 2008.

56. Thurston WE, Coupal S, Jones CA, Crowshoe LF, Marshall DA, Homik J, et al. Discordant indigenous and provider frames explain challenges in improving access to arthritis care: A qualitative study using constructivist grounded theory. Int J Equity Health 2014;13:46.

57. Kottak N, Tesser J, Leibowitz E, Rosenberg M, Parenti D, DeHoratius R. Ethnographic observational study of the biologic initiation conversation between rheumatologists and biologic-naive rheumatoid arthritis patients. Arthritis Care Res (Hoboken) 2018;70:997-1004.

58. Rose G. Why do patients with rheumatoid arthritis use complementary therapies? Musculoskeletal Care 2006;4:101-15.

59. Grant M. Mothers with arthritis, child care and occupational therapy: Insight through case studies. Br J Occup Ther 2001;64:322-9.

60. Stamm T, Lovelock L, Stew G, Nell V, Smolen J, Machold K, et al. I have a disease but i am not ill: A narrative study of occupational balance in people with rheumatoid arthritis. OTJR Occup Particip Health 2009;29:32-9.

61. Kuper A, Lingard L, Levinson W. Critically appraising qualitative research. BMJ 2008;337:a1035.

62. Morse J. Determining sample size. Qual Health Res 2000;10:3-5.

63. Morgan DL. Focus groups as qualitative research. 2nd ed. Thousand Oaks CA: Sage publications; 1996.

64. Pope C, Mays N. Qualitative research in health care. 3rd ed. Boston: Blackwell Publishing [BMJ books]; 2006.

65. Bowen GA. Document analysis as a qualitative research method. Qual Res J 2009;9:27-40.

66. Braun V, Clarke V, Hayfield N, Terry G. Thematic analysis. In: Liamputtong P, editor. Handbook of research methods in health social sciences. Singapore: Springer; 2019. p. 843-60.

67. Reeves S, Albert M, Kuper A, Hodges BD. Why use theories in qualitative research? BMJ 2008;337:a949.

68. Creswell JW, Creswell JD. Mixed methods procedures. In: Salmon h, neve c, felts dc, marks a, editors. Research design: Qualitative, quantitative, and mixed methods approaches. Los angeles, sage publications; 2018:213-46.

69. Kelly A, Tymms K, de Wit M, Bartlett SJ, Cross M, Dawson T, et al. Patient and caregiver priorities for medication adherence in gout, osteoporosis and rheumatoid arthritis: Nominal group technique. Arthritis Care Res 2019 Jul 19 [E-pub ahead of print].

70. Tunnicliffe DJ, Singh-Grewal D, Craig JC, Howell M, Tugwell P, Mackie F, et al. Healthcare and research priorities of adolescents and young adults with systemic lupus erythematosus: A mixed-methods study. J Rheumatol 2017;44:444-51.

71. Carr ECJ, Ortiz MM, Patel JN, Barber CEH, Katz S, Robert J, et al. Models of arthritis care: A systems-level evaluation of acceptability as a dimension of quality of care. J Rheumatol 2019 Nov 15 (E-pub ahead of print).

72. Mahmood S, Hazes JMW, Veldt P, van Riel P, Landewé R, Bernelot Moens H, et al. The development and evaluation of personalized training in shared decision-making skills for rheumatologists. J Rheumatol 2020;47:290-7.

73. Nicklin J, Cramp F, Kirwan J, Greenwood R, Urban M, Hewlett S. Measuring fatigue in rheumatoid arthritis: A cross-sectional study to evaluate the bristol rheumatoid arthritis fatigue multi-dimensional questionnaire, visual analog scales, and numerical rating scales. Arthritis Care Res (Hoboken) 2010;62:1559-68.

74. Dures EK, Hewlett SE, Cramp FA, Greenwood R, Nicklin JK, Urban M, et al. Reliability and sensitivity to change of the bristol rheumatoid arthritis fatigue scales. Rheumatology (Oxford) 2013;52:1832-9.

75. Lewin S, Glenton C, Oxman AD. Use of qualitative methods alongside randomised controlled trials of complex healthcare interventions: Methodological study. BMJ 2009;339:b3496.

76. Onwuegbuzie AJ, Leech NL. Linking research questions to mixed methods data analysis procedures 1. Qual Report 2006;11:474-98.

77. Oakley A, Strange V, Bonell C, Allen E, Stephenson J. Process evaluation in randomised controlled trials of complex interventions. BMJ 2006;332:413-6.

78. Flurey CA, Hewlett S, Rodham K, White A, Noddings R, Kirwan JR. Identifying different typologies of experiences and coping strategies in men with rheumatoid arthritis: A q-methodology study. BMJ Open 2016;6:e012051.

79. Blasco-Blasco M, Ruiz-Cantero MT, Juarez-Herrera YCLA, Jovani V, Pascual E. Sex and gender interactions in the lives of patients with spondyloarthritis in spain: A quantitative-qualitative study. J Rheumatol 2017;44:1429-35.

80. Singh JA, Herbey I, Bharat A, Dinnella JE, Pullman-Mooar S, Eisen S, et al. Gout self-management in african american veterans: A qualitative exploration of challenges and solutions from patients' perspectives. Arthritis Care Res 2017;69:1724-32.

81. Craig ET, Orbai AM, Mackie S, Bartlett SJ, Bingham CO, 3rd, Goodman S, et al. Advancing stiffness measurement in rheumatic disease: Report from the stiffness special interest group at omeract 2018. J Rheumatol 2019;46:1374-8.

82. Lima K, Phillip CR, Williams J, Peterson J, Feldman CH, Ramsey-Goldman R. Factors associated with participation in rheumatic disease-related research among underrepresented populations: A qualitative systematic review. Arthritis Care Res 2019.

83. Karp DR, Chong BF, James JA, Arriens C, Ishimori M, Wallace DJ, et al. Mock recruitment for the study of antimalarials in incomplete lupus erythematosus trial. Arthritis Care Res 2019;71:1425-9.

84. Lawford BJ, Delany C, Bennell KL, Hinman RS. "I was really pleasantly surprised": Firsthand experience and shifts in physical therapist perceptions of telephone-delivered exercise therapy for knee osteoarthritis-a qualitative study. Arthritis Care Res 2019;71:545-57.

85. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (coreq): A 32-item checklist for interviews and focus groups. Int J Qual Health Care 2007;19:349-57.

86. Critical appraisal skills programme (2018). Casp qualitative checklist.; [cited 12/09/2019]; Available from: https://casp-uk.net/casp-tools-checklists/.

87. Guba EG. Criteria for assessing the trustworthiness of naturalistic inquiries. Educ Tech Res Develop 1981;29:75-91.

88. Levitt HM, Bamberg M, Creswell JW, Frost DM, Josselson R, Suárez-Orozco C. Journal article reporting standards for qualitative primary, qualitative meta-analytic, and mixed methods research in psychology: The apa publications and communications board task force report. Am Psychol 2018;73:26-46.

89. Greenhalgh T, Taylor R. Papers that go beyond numbers (qualitative research). BMJ 1997;315:740-3.

90. Sandelowski M. Rigor or rigor mortis: The problem of rigor in qualitative research revisited. ANS Adv Nurs Sci 1993;16:1-8.

91. Lincoln YS, Guba EG. Naturalistic inquiry. Newberry park. London, UK: Sage Publications; 1985.

Table 1 Selected examples of qualitative studies in rheumatology

Reference	Topic	Approach	Data collection	Data analysis	Main findings	Implications for clinical care or policy
Singh et al. (13)	Gout self-management among African American male veterans with high medication adherence	NR	Semi-structured interviews	Thematic analysis and content analysis	Fear of the severe and debilitating pain of gout and self-confidence from having a military-like discipline helped veterans adhere to medications and lifestyle changes. Maintaining a positive outlook and accepting the diagnosis of gout allowed participants to embrace changes in their daily routines.	Emphasizing self-discipline, positivity and disease acceptance through patient narratives could facilitate better gout-self management.
Flurey et al. (14)	Experiences and coping styles of men with RA	NR	Focus groups	Thematic analysis	Men with RA felt angry, embarrassed and helpless by their reduction in strength, ability to work, perform household duties or play with children. Participants coped by being stoic or reacted by engaging in destructive behaviours, withdrawing and concealing their arthritis.	Health professionals should ask men explicitly about their psychological and emotional well-being and be aware that RA can threaten masculine roles and identity. Support could take the form of purposeful information-oriented sessions with other patients with RA.
Hart et al. (15)	How young people (aged 16-25) with inflammatory arthritis evaluate the risks and benefits of treatment	Grounded theory	Semi-structured interviews, recorded consultations, focus groups	Grounded theory analysis	Young people aspired to live a "normal" life. However, treatment schedules and side effects could be highly intrusive, diminish well-being, and compound feelings of being different. Changes to treatment could force young people to confront their illness and heighten distress about uncertainties of the future. Participants wished for a relatively simple and stable treatment regimen that improved symptoms and had side effects that posed minimal restrictions.	Health professionals should elicit young people's priorities and concerns regarding their treatment and address the impact of treatment on their ideas of a "normal" life such as relationships, education, work and physical appearance.
Shaw et al. (16)	The development of resilience among patients with RA	Ethnography/ Narrative inquiry	Semi-structured interviews and observations of participants' living environment and routines	Narrative analysis	Resilience was cultivated through internally directed emotional management strategies and externally directed behaviours. This included adopting a mindset of being in control of their RA, remaining calm during challenges such as medication-related complications, and positive reframing and focus on abilities rather than limitations. Offering support to others through community service, engaging in enjoyable hobbies and activities, or using humour in social interactions about limitations posed from RA allowed patients to feel valuable, satisfied and connected to others.	Behavioural interventions or social support programs can promote resilience by utilizing externally and internally directed management strategies identified in the study.

Reference	Topic	Approach	Data collection	Data analysis	Main findings	Implications for clinical care or policy
Tunnicliffe et al. (17)	Healthcare and research priorities of adolescent and young adults with SLE	Mixed methods study	Semi-structured interviews and focus groups	Thematic analysis, descriptive statistics for votes	Service shortfalls including timely diagnosis of SLE worsened symptoms and caused anxiety, lack of culturally relevant educational materials made it difficult for participants to understand and explain their illness to family and friends. Participants strongly emphasized the impact of SLE on psychological health including reduced self-esteem, social withdrawal and fear of being unable to achieve future vocational and family goals. Participants also wanted to reduce the psychological, emotional and financial burden SLE imposed on their family, friends and other patients with life disrupting manifestations of SLE.	Research and clinical resource allocation should address gaps in service provision and incorporate strategies to alleviate anxiety and efficient use of resources to minimize the impact of SLE on family, friends as well as the wider population of patients with SLE.
Sumpton et al. (18)	Patients' perspectives of systemic sclerosis	NR	Semi-structured interviews	Thematic analysis	Systemic sclerosis imposes major physical and social restrictions that impair patients' identity and self-esteem. Insecurities and anxiety in care arise from ambiguities about the cause, diagnosis and prognosis of the disease.	Clinical care for patients with systemic sclerosis can be optimized by providing psychosocial care and improving communication and education around the concerns regarding disease prognosis and management.
Hewlett et al. (19)	RA patients' perspectives of flare	NR	Focus groups	Thematic analysis	Flare incorporated an individual cluster of symptoms including severe, unrelenting and multi-joint pain, dramatic and extreme level of stiffness, fatigue that was unlike normal RA fatigue, systemic flu-like symptoms and cognitive shut down. The symptoms profoundly compromised simple daily functions and caused emotional distress to the point of wanting to cut off joints or die for some. Patients increased their usual level of self-management and would seek professional help when they were still unable to control their multiple symptoms or run their normal lives.	Assessment of flare that includes these patient experiences can help patients and clinicians recognize early warning signs and enhance communication between patients and professionals.

NR, Not reported; RA, Rheumatoid arthritis; SLE, Systemic lupus erythematosus

Table 2. Appraisal of qualitative studies

Qualitative criteria	Quantitative criteria	Aspect of quality	Examples from COREQ domains and items
Credibility: Are the findings trustworthy?	Internal validity	Truth value	Purposive sampling Data saturation Final sample size Interview guide Duration Repeat interviews Setting of data collection Relationship with participant Presence of non-participants Multiple data coders Participant checking Clarity of themes
Confirmability: Are the findings linked to the data?	Objectivity	Neutrality	Reflexivity Multiple data coders Participant checking Quotations Data and findings consistent
Dependability: Is the process auditable?	Reliability	Consistency	Audio/Visual recording Transcription Description of coding tree Software
Transferability: Are the findings relevant to other contexts?	Generalisability	Applicability	Thick description of sample and setting of data collection

Table 2. Appraisal of qualitative studies using the Lincoln and Guba framework linked to examples from Consolidated Criteria for Reporting Qualitative Health Research (COREQ) items.

	Description	Participant selection	Data collection	Data analysis	
Grounded theory	Develops a theory of a process or action grounded in the data	Theoretical sampling	Typically interviews	Iterative data collection, analysis and memoing (of ideas). Constant comparison (of collected data with emerging theories). Can be structured (e.g. open, axial and selective coding).	Health care access of Aboriginal people with arthritis (56)
Ethnography	Describes shared patterns of behaviours, beliefs, language of a sociocultural group	Individuals in a socio-cultural group, often via gatekeepers/key informants	Extensive fieldwork, typically involving observations and interviews	Description of the socio-cultural group, thematic analysis of how the group works and lives, and an overall picture of how the system works.	Physician-patient biologic initiation conversations (57)
Phenomenology	Identifies the essence of the lived human experience of a phenomenon	Several individuals with the lived experience of a phenomenon	Typically in-depth and repeated individual interviews	Narrow units (significant statements), broader units (meaning clusters), then detailed description of the experience and essence of the phenomenon.	Complementary therapies in rheumatoid arthritis (58)
Case studies	Detailed exploration of one or more cases (e.g. individuals, programs)	Typically current, real-life case(s) within a specific time and place	Multiple forms of data (e.g. interviews, observations, documents)	Single or multiple case analysis. Case description and case themes.	Occupational therapy in mothers with arthritis (59)
Narrative research	Tells the stories of one or more individuals	One or more individuals who have life experiences or stories to share	In-depth interviews and other data (e.g. diaries, letters, documents)	Chronological restorying (reorganising). Context of culture/history provided. Collaborative approach with participant.	Engagement in occupations in rheumatoid arthritis (60)