

Do patients with rheumatic diseases living in rural settings have poorer outcomes? Results from a systematic review of the literature

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Abstract

Objectives: To assess whether clinical and patient-reported outcomes are poorer for people with inflammatory and non-inflammatory rheumatic diseases living in rural locations.

Methods: We searched six databases for articles which were primary peer-reviewed research, published in English 1990-2019, which focussed on selected rheumatic diseases (rheumatoid arthritis (RA), psoriatic arthritis (PsA), axial spondyloarthritis (axSpA) or osteoarthritis (OA)), and quantified either patient-reported or clinically measured outcomes by a measure of rurality or remoteness. Selected articles were synthesised narratively.

Results: Eight eligible publications, including 753 rural and 929 urban patients, evaluated outcomes in RA (5 studies) and OA (3 studies). Studies were small, single centre, and rarely provided a definition of rurality. Aspects relating to rurality, such as access to services, were not measured. In RA some studies suggested greater functional disability and disease activity in rural dwellers. In OA, there was some evidence to suggest that rural dwellers presented with more advanced degenerative hip changes, and that illness perceptions and coping differed between rural and urban dwellers. No studies examined work outcomes. Potentially important confounding factors such as socio-economic status were rarely considered.

Conclusion: There remains considerable uncertainty whether outcomes differ for rheumatic disease patients in rural settings. There is a need for larger-scale studies characterising participants in relation to place of residence, in order to determine whether rurality is an independent predictor of outcome or a surrogate marker for socioeconomic factors.

Significance and Innovation

- Few and poor quality studies have examined treatment outcomes in patients with rheumatic disease living in rural settings.
- Rurality (and related factors) are poorly defined in studies which have been conducted.
- The role of potentially important confounding factors e.g. social and economic factors has not been considered.
- Mixed-methods approaches are necessary to understand the complex interplay between rurality and health outcomes.

Equitable and timely access to specialist, multi-disciplinary care and support for those with rheumatic disease is essential to prevent poor outcomes such as joint deformities, functional limitations and disability (1). Most specialist services are located in urban areas, yet the ageing, multi-morbid population is increasing faster in rural areas (2). Geographical location has a significant impact on health inequalities, with social exclusion and isolation, access to and awareness of health services, poor housing, low income, travel distance, availability and accessibility of transport impacting disproportionately on rural communities (2).

Delivering and sustaining quality healthcare services to rural areas to manage the needs of patients with chronic, complex conditions is challenging; for example, recruiting and retaining an appropriately skilled workforce, and difficulties realising economies of scale while adequately serving sparsely populated areas (2). Many studies have highlighted inadequate access to specialist healthcare services for those with rheumatic and musculoskeletal disease (RMD) living in rural and remote locations (3). However, whether patients with RMD living in rural settings have poorer outcomes remains unknown and there has been, as far as we are aware, no review of the evidence.

This systematic review aims to assess whether clinical and patient-reported outcomes are poorer for people with inflammatory and non-inflammatory rheumatic diseases living in rural locations.

Materials and Methods

Literature search

MEDLINE, EMBASE, CINAHL, PsycINFO, Web of Science and Cochrane Library were searched using Medical Subject Headings (MeSH) and keywords spanning the following fields: selected rheumatic diseases (rheumatoid arthritis, psoriatic arthritis, axial spondyloarthritis/ankylosing spondylitis and osteoarthritis), rural or urban area of residence and disease outcomes. The latter included clinical status/disease activity, patient or physician global assessment (including quality of life) and measures of function. The search strategy was initially developed for MEDLINE and adapted for each database.

Study eligibility

Publications were eligible for inclusion if they met the following criteria: the publication (1) reported primary research and was in a peer-reviewed journal; (2) focused on adult patients with rheumatoid arthritis (RA), psoriatic arthritis (PsA), axial spondyloarthritis/ankylosing spondylitis (axSpA) and/or osteoarthritis (OA), or these patient groups could be separately identified (3) compared and quantified at least one measure of disease outcome (clinical or patient reported) in patients resident in rural and urban areas and (5) was published in English between January 1990 and July 2019. We chose 1990 as the earliest date since studies

conducted prior to this would have involved considerably different approaches to the clinical management of eligible patients.

Study selection

After duplicate removal, one reviewer screened all records by title, abstract and subsequently full-text to determine inclusion. Uncertainties were resolved by consensus. Bibliographies of all included publications were manually searched to obtain additional relevant publications. Relevant data were extracted by one reviewer and checked by a second reviewer.

Due to the heterogeneity of study design, diseases investigated and outcomes measured, a meta-analysis was not conducted. Data were extracted and summarised narratively.

Results

A total of 8 publications were identified as including an eligible study. No additional publications were included after screening their reference lists (Figure 1). Eligible studies included a total of 753 and 929 patients in rural and urban locations respectively from 8 countries (2 from Europe, 3 Asia, 2 Africa and 1 from the Americas). Of the eligible studies, 5 investigated RA, 3 studies OA (1 hip OA, 2 knee OA); there were no studies identified on PsA or axSpA. With the exception of one study which was longitudinal in design, all others were cross-sectional. Most studies examined patients attending specialist rheumatology or orthopaedic services (Table 1).

In describing studies, focus has principally been on recognised measures of clinical status (e.g. disease activity or degree of degeneration), patient/clinician global assessment of disease (including quality of life), measures of function and work productivity. Specifically we also note the definition used (if any) of rural areas and whether comparisons between populations are adjusted for factors which could potentially confound the relationship, namely demographic and socioeconomic factors.

Rheumatoid Arthritis

Puchner et al (2014) (4) enrolled rheumatologists, primarily, whose practice involved rural patients across three provinces of Austria. They provided a questionnaire which was distributed to consecutive patients with RA which were then completed at home. In terms of rurality this was measured by three parameters a) size of settlement in which they lived b) time to travel to provincial capital and c) time to travel to their rheumatologist. Of 124 participants, 103 described that they lived in a settlement of less than 50,000 persons. There were no differences in patient reported health status according to any of the above measures of rurality.

Lekpa et al (2012) (5) recruited patients with RA (according to American College of Rheumatology (ACR) 1987 criteria) from the rheumatology outpatient department in Dakar,

Senegal. The primary purpose of the report was to compare urban and rural patients. Urban patients were defined as those resident in the capital city or in an administrative centre (“chef-lieu”) of the region, while all other patients were classified as living in rural settings. The study included 180 patients of which 143 (79%) lived in urban areas. Comparing the two groups, there were no differences in the presence or type of hand deformities. In those from rural areas, the median level of swollen joints (6 v 4) and DAS-28 (7.2 v 6.4) was higher although neither difference was statistically significant. A greater proportion of rural patients had extra-articular manifestations (70.3% vs 49%). There was no difference in the presence of rheumatoid factor nor anti-citrullinated protein antibody (ACPA) between the two groups. There was no account of confounding factors made in comparing the groups, but it was noted that there were marked gender differences, with men more likely to be from rural areas than women (41% v. 18%).

Zhao et al (2015) (6) recruited 607 patients meeting 1987 ACR criteria for RA from a rheumatology outpatient department in Chengdu, China. Residence was categorised as urban (n=222), suburban (n=116) or rural (n=269), although no details were provided about how this classification was made. Clinical and self-reported information was collected and related to the Health Assessment Questionnaire Disability Index (HAQ-DI). Functional disability significantly increased across people living in urban, suburban and rural settings. However, this study additionally performed a multivariable analysis in which living in a rural setting was an independent predictor of increased functional disability (equivalent to a 1.23 point increase in HAQ-DI). Other independent predictors were lack of available social support, older age, pain, number of times hospitalised and disease duration [note this interpretation is from the tables as the text in this study gives contradictory interpretation of the data]. Across the study population, 70% were educated below junior high school and 40% had a monthly household income monthly per capita < \$160. However, education level and household income was not adjusted for in the analysis.

Alarcón et al (2015) (7) recruited 189 patients from the rheumatology clinic of a referral centre in one region of Chile. It also acted as a referral centre for other regions. Rural residence was defined as “living in the scattered agricultural communities” and 61 participants were classified as such. Disability was measured using the Spanish version of the HAQ and dichotomised into moderate/severe v. slight, due to sparse data. Rural residence was associated with moderate/severe disability (Odds Ratio (OR_{crude}) 3.3, 95% CI (1.2, 11.6)) but with considerable uncertainty around the level of association. On multivariable analysis the strength of association reduced and was not significant, OR_{adj} 2.1, 95% CI (0.6, 7.1) after adjusting for socio-economic status, ethnicity, gender and age. This study was probably too small to be able to conduct a robust multivariable analysis.

In a study which involved comparing black Zimbabwean patients and Caucasian UK patients with RA (according to the American Rheumatism Association 1958 criteria), Chikanza et al, 1994 (8) presented data comparing urban (n=41) and rural (n=43) Zimbabwean participants attending a tertiary rheumatology clinic. No definition of how rural status was defined was

included in the report. Across a variety of clinical, serological and radiological measures of RA the data show no differences of large magnitude and the text reports no significant differences.

Osteoarthritis

Roopsawang and Aree-Ue (2015) (9) undertook an interview survey in three communities in Bangkok (Thailand) and three communities in its vicinity. Participants were recruited through health centres and community leaders and were considered to have knee osteoarthritis based on symptoms and signs using ACR criteria. There were 116 and 112 participants classified as rural and urban respectively but there were no details given on the classification. There were no confounding factors considered in comparisons but it was noted that those in rural areas were considerably more likely to be of normal weight (35% v. 19%) and less likely to have co-morbidities (35% v. 47%). Only 5.2% of rural dwellers were educated to high school level compared to 34.8% of urban dwellers. This study only reported on illness representation and coping behaviour. Rural dwellers were less likely to perceive their symptoms as curable, more likely to use spiritual coping methods and less likely to use cognitive-focused coping behaviours e.g. information seeking and self-care. Those in rural areas reported a lower level of emotional impact of symptoms.

Rapała et al (2015) (10) reported on 200 patients who were about to undergo total hip arthroplasty for osteoarthritis. There were 79 and 121 patients classified as rural and urban respectively but there was no indication of how the classification was made. In terms of clinical status, using a scale proposed by Garlicki and Kreczka patients from rural settings were much more likely to have the most advanced level of hip degenerative changes on the three point scale “almost 70% of ...rural patients ...compared to only 44.5% of urban patients”. No account was taken of potential confounding factors but data presented demonstrated no large differences in mean age, proportion male and mean Body Mass Index (BMI) between those classified as urban and rural.

Çankaya et al (2016) (11) followed 70 patients prospectively with unilateral primary knee osteoarthritis who were undergoing arthroplasty. Of these, 45 were classified as rural but no information was given on the classification. Outcome was measured six months post-surgery by functional status using the Knee Injury and Osteoarthritis Outcome Score Physical Function Short Form (KOOS-PS). Quality of life was assessed using the Short-Form 36. There was no important or statistically significant difference in change in function or quality of life comparing those resident in rural and urban settings. No effect of potential confounding was considered (all analyses considered only individual variables) but the study did demonstrate that benefit (in terms of quality of life) did increase with higher levels of education and absence of other co-morbidities.

Discussion

We have identified only a small number of studies examining clinical or patient reported measures of outcome in rheumatoid arthritis and osteoarthritis, and none in psoriatic arthritis and axial spondyloarthritis. All studies are relatively small, rarely provide a definition of rural status (and specifically do not measure aspects related to rurality such as access to services), and do not consider factors which could confound any differences observed between urban and rural patients. No eligible studies examined work outcomes between urban and rural patients.

The aim of this systematic review was to determine whether clinical and patient-reported outcomes were poorer for people with inflammatory and non-inflammatory rheumatic diseases living in rural locations. We excluded papers that did not differentiate between specific rheumatic diseases. Inflammatory and non-inflammatory rheumatic disease have different underlying pathophysiology, service delivery and resource requirements. To help inform the type of approaches required to reduce any inequalities, comparisons therefore need to be precise; any observed differences in outcomes are only meaningful if differences between and across conditions can be determined. For example, are differences in outcome driven by specific conditions, or common across all conditions? However, we did identify two large studies from the US (12) and Australia (13) which, although did not differentiate between types of arthritis, were otherwise relevant. Kovac et al (12) examined health-related quality of life among 1,191 individuals with self-reported arthritis (mainly RA and OA). After adjusting for socio-economic status, rural residency was an independent predictor of poorer physical and mental health. Dowsey et al (13) found that rural patients in Australia presented at a younger age for hip and knee replacement, and with less severe radiographic disease. The authors postulate this may reflect rural dwellers working in physical jobs seeking referral earlier due to concerns about disease progression, and lack of multi-disciplinary support for self-management in rural communities. In contrast, Rapala et al, 2005 (10) found that rural patients in Poland presented with more severe degenerative joint disease. This suggests potential differences in illness perceptions, health seeking behaviour and coping mechanisms between urban and rural dwellers with arthritis across different healthcare contexts and cultures.

Our systematic literature review has several limitations. Data was scarce and most studies only sampled rural patients attending urban-based specialist centres; this is likely to underestimate any geographical differences. The term “rurality” can encompass many relevant issues in terms of health and health care including; population density, population size (which are related to rurality), and availability of and travel time to healthcare services (more related to remoteness). Only one included study attempted to measure more than one of these aspects – most relied on a single geographical descriptor or did not provide any definition of rurality. Studies were conducted across diverse healthcare contexts, with significant variation in access and provision of specialist RMD services, and payment systems.

However, these important contextual factors and their potential influence on access to services for those living in rural areas were not measured, making it difficult to draw comparisons and explore reasons for any disparities between urban and rural dwellers. Most studies were cross-sectional which precluded examination of changes over time or response to therapy.

Low socioeconomic status has been associated with worse clinical outcomes, decreased functional ability and reduced quality of life in RA (14). Whilst rural areas in developed countries are often considered to be less deprived, 'hidden' rural deprivation is increasingly recognised, representing a complex interplay between factors associated with income, social circumstances, access to services, and patient choice that is not captured by existing area-based measurements of deprivation (15). Whilst several studies in this review reported lower education and income levels in rural settings, most eligible studies did not adjust for socio-economic status. Those that did (7) were underpowered to examine multivariable relationships. It is therefore difficult to determine, based on existing evidence, whether rurality is an independent predictor of poor outcome in RMD or a surrogate marker for socio-economic status. We did not undertake a formal quality assessment as we had already identified that almost all studies failed in terms of two key issues: the definition of rural settings and taking account of confounding factors when examining the relationship between residence and outcome.

In conclusion, we have identified key priorities for future research. Studies using population level data are necessary to capture the burden of disease and health outcomes in RMD between rural and urban areas. Differentiating between conditions is an important point to be considered when designing future research to examine rural-urban differences in outcomes in rheumatic disease. There is also a need to define rurality consistently to allow comparison across studies, and have valid and measurable indicators of rural deprivation to explore the independent effect of rurality on health outcomes. Mixed-methods approaches provide additional opportunities to explore the complex interplay between rurality and health outcomes in RMD.

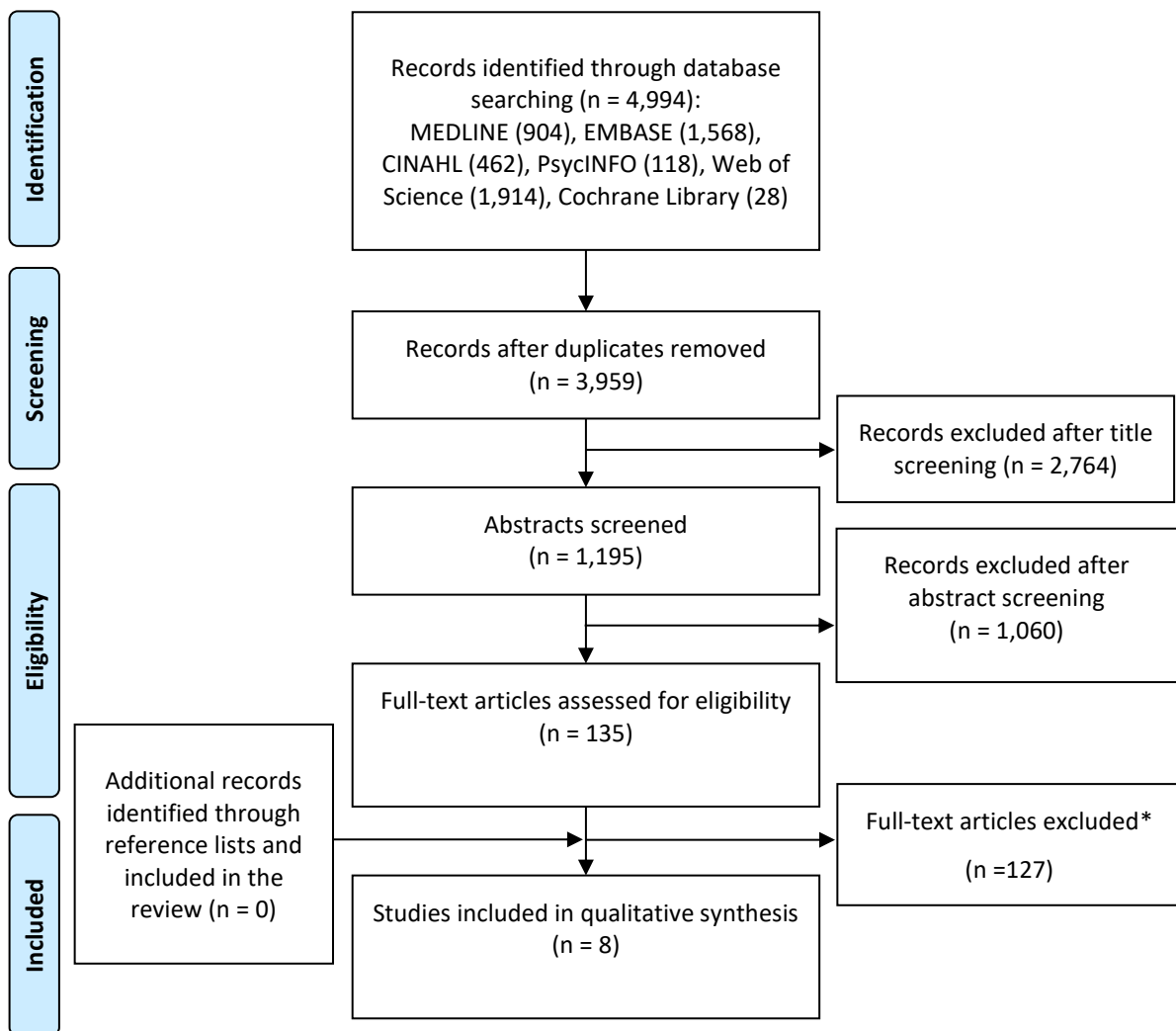
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Figure 1: Flow chart identifying eligible studies



* Exclusion of full-text articles due to the following reasons: Full-text not English (n = 1), conference abstract only (n=9), no primary peer-reviewed study (n=10), not investigating disease of interest/did not differentiate between different types of arthritis (n=29), no comparison of rural and urban patients (n =44), not investigating disease outcomes (n=34).