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Multicriteria decision analysis methods with 1000Minds for developing systemic sclerosis classification criteria

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Abstract

Objectives: Classification criteria for systemic sclerosis (SSc) are being developed. The objectives were to develop an instrument for collating case data and evaluate its sensibility; use forced-choice methods to reduce and weight criteria; and explore agreement among experts on the probability that cases were classified as SSc.

Study Design and Setting: A standardized instrument was tested for sensibility. The instrument was applied to 20 cases covering a range of probabilities that each had SSc. Experts rank ordered cases from highest to lowest probability; reduced and weighted the criteria using forced-choice methods; and reranked the cases. Consistency in rankings was evaluated using intraclass correlation coefficients (ICCs).

Results: Experts endorsed clarity (83%), comprehensibility (100%), face and content validity (100%). Criteria were weighted (points): finger skin thickening (14−22), fingertip lesions (9−21), friction rubs (21), finger flexion contractures (16), pulmonary fibrosis (14), SSc-related antibodies (15), Raynaud phenomenon (13), calcinosis (12), pulmonary hypertension (11), renal crisis (11), telangiectasia (10), abnormal nailfold capillaries (10), esophageal dilation (7), and puffy fingers (5). The ICC across experts was 0.73 [95% confidence interval (CI): 0.58, 0.86] and improved to 0.80 (95% CI: 0.68, 0.90).

Conclusions: Using a sensible instrument and forced-choice methods, the number of criteria were reduced by 39% (range, 23−14) and weighted. Our methods reflect the rigors of measurement science and serve as a template for developing classification criteria.

Keywords: Scleroderma; Systemic sclerosis; Decision analysis; Forced-choice; Classification criteria; Conjoint analysis; Sensibility

1. Introduction

Classification criteria for rheumatic diseases are important for research and practice. Previous iterations of classification criteria have been criticized for their lack of methodological rigor, inability to reflect the changing construct of disease, or inefficiency when applied in the real world [1−4]. Application of the preliminary criteria for classification of systemic sclerosis (SSc) [5−7] for recruitment into trials results in the exclusion of approximately 20% of patients with either early SSc or the limited subtype [8−10]. As a result, new classification criteria for SSc are being developed [11].

In phase 1, a total of 168 candidate criteria were generated through Delphi exercises [11]. The items were reduced to 23 criteria using another Delphi exercise and nominal group technique. The 23 criteria were validated using SSc and SSc-mimicking condition cohorts. The criteria were found to have good face, discriminant, and construct validity [12]. The next phase of criteria development requires further item reduction, weighting, and scaling. The 2010 rheumatoid arthritis classification criteria were successfully developed with a balanced use of expert-based and data-driven methods. Forced-choice methods (facilitated by 1000Minds software; http://www.1000Minds.com) allowed for item reduction and item weighting [13,14]. To use these methods in the development of SSc classification criteria, an SSc-specific instrument using a standardized format based on the 23 candidate items needed to be developed [15].

The aim of this study was to develop an SSc-specific instrument for use in the forced-choice phase of SSc criteria development and conduct a forced-choice study to reduce and weight the criteria. The sensibility of an instrument is critical to whether it is useful or not [15]. Attributes of sensibility include comprehensibility, clarity, face validity, content validity, and feasibility. If an instrument is not sensible, it does not warrant use in clinical research [15]. Therefore, the first objective was to develop and evaluate the sensibility of an SSc-specific instrument for use in a forced-choice study. The second objective was to use forced-choice methods to reduce the criteria and valuate relative weights for each criterion. The third objective was to explore the agreement among experts on which patients are considered to have SSc. Given the heterogeneity of SSc, we hypothesized that in the absence of distal AND proximal skin thickening present at the same time, there would be variability in agreement among experts on which patients have SSc. If this hypothesis were true, it would provide justification for the need to apply standardized classification criteria for inclusion of patients into research studies.

2. Methods

2.1. Candidate criteria

Items were generated through consensus exercises resulting in 168 candidate criteria. A Delphi exercise and nominal group technique reduced the candidate criteria to 23:

1. antinuclear antibody;
2. anti−topoisomerase-I antibody;
3. anticentromere antibody or centromere pattern on antinuclear antibody test;
4. anti-RNA polymerase III antibody;
5. anti-PM-Scl antibody;
6. scleroderma;
7. puffy fingers;
8. finger flexion contractures;