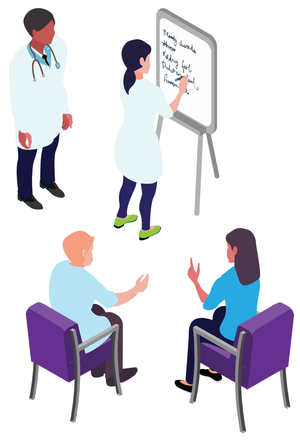
Authoring of Peer-Reviewed Articles on the Experiences of Patients with Rare Diseases by Patients and their Caregivers: A Rapid Review

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Background

- Through lived experience, patients and caregivers can provide unique insight into disease signs, symptoms, course, burden, and treatment needs.[1]
- This is especially the case in rare diseases, where frequently little information is available due to low prevalence.[2,3]
- Partnering with patients and caregivers as co-authors of peer-reviewed articles is increasing and makes sense, especially for articles describing patient experiences.[4-6]

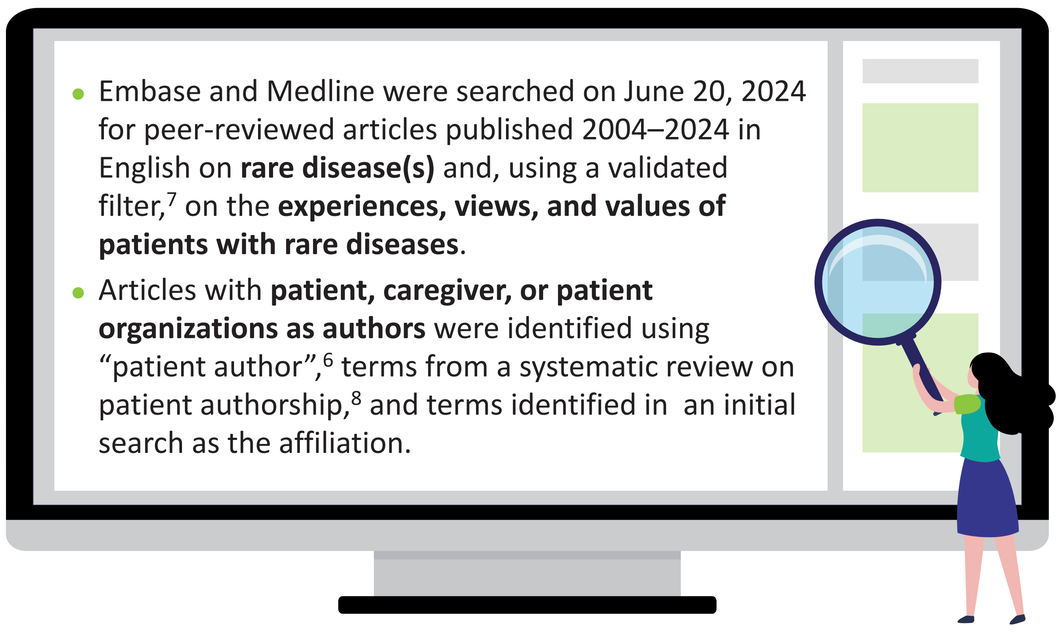


Objective

Examine the practice of including patients and caregivers as byline authors of peer-reviewed articles on the experiences of patients with rare diseases.

Methods

- Embase and Medline were searched on June 20, 2024 for peer-reviewed articles published 2004–2024 in English on **rare disease(s)** and, using a validated filter,[7] on the **experiences, views, and values of patients with rare diseases**.
- Articles with **patient, caregiver, or patient organizations as authors** were identified using "patient author",[6] terms from a systematic review on patient authorship,[8] and terms identified in an initial search as the affiliation.



Results

Sample

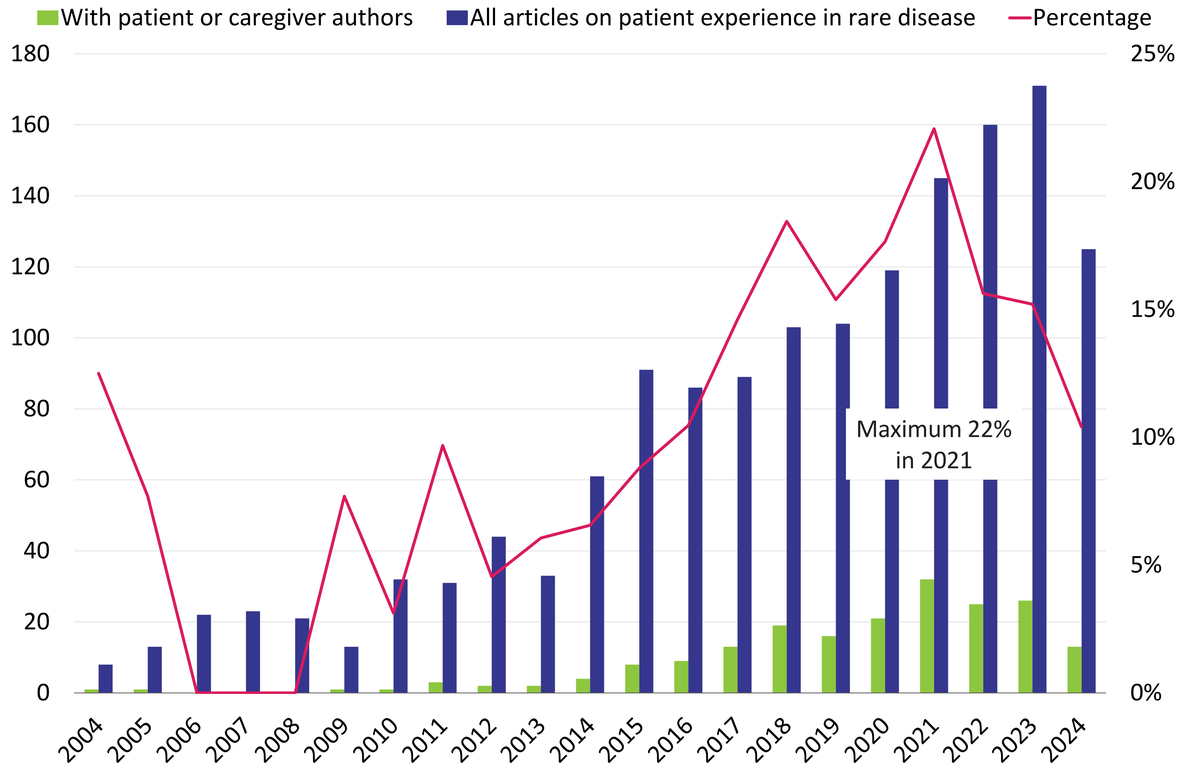
1494
Total articles selected

197
Articles with patient or caregiver authors

Extensive manual work was needed to eliminate records with irrelevant affiliations.

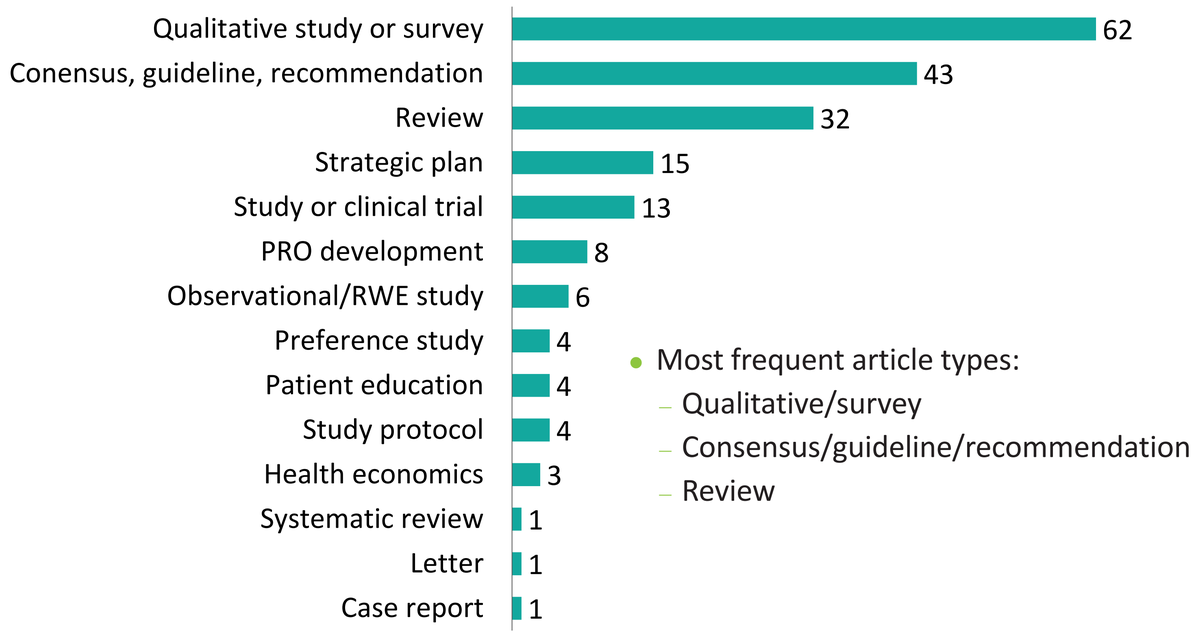
- Numbers increased steadily since 2004 (**Figure 1**)
- Proportion with patient/caregiver authors increased steadily with time to 22% in 2021 but has fallen steadily since

Figure 1. Articles Per Year



Results (cont.)

Figure 2. Article Type



- Most frequent article types:
  - Qualitative/survey
  - Consensus/guideline/recommendation
  - Review

Abbreviations: PRO=patient-reported outcome; RWE=real-world evidence

Figure 3. Affiliation Terms



- Associations were the identifying affiliation for 95%
  - This makes it unclear whether the author(s) themselves had lived experience.
- "Patient author", promoted recently as an affiliation,[8] was listed for only a single article.
- A patient, caregiver, or patient association was the first author for 22% and the only author for 7%.

Conclusions

- Patients, caregivers, and especially representatives of patient associations are increasingly visible as co-authors of peer-reviewed articles on the experiences, views, and values of patients with rare diseases.
- Patients and caregiver partners in research can — and should have the opportunity to — be authors of peer-reviewed articles in accordance with authorship guidelines.
- To better understand the role and impact of people with lived experience as authors, we need consistent ways of describing their participation and identifying them in publication databases that also respect their preferences and privacy concerns.

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