

Who's Who in Rare Diseases: A Case Study in Author Identification

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Objective

- Using phenylketonuria as a case study, we aimed to identify relevant key opinion leaders by quantifying previous authorship contributions and exploring prior collaborations between experts.

Background

- When publishing in an unfamiliar therapy area, it is important for researchers to identify key opinion leaders in order to elicit expert opinion, thereby establishing a clear and up-to-date understanding of an indication or treatment.
- This is particularly challenging in rare diseases, which are typically multi-system conditions with a wide range of symptoms. Consequently, there is often a need to identify a broad range of specialists involved in management of rare conditions, with only a limited number of disease-specific experts available.
- A method to identify the most prominent key opinion leaders in a rare disease field, in addition to the experts they most commonly collaborate with, would be valuable for professionals aiming to publish in this area.

Methods

- Abstracts with the term "phenylketonuria" in their title or body text from 01/01/2016–19/09/2018 were identified via PubMed using a pragmatic literature search.
- Different variations of author names were assessed for spelling differences and standardised as required. Duplicate publications were removed.
- Authors in the by line of ≥ 1 publication were selected and stratified according to number of phenylketonuria publications on which they had been listed as a (first) author using R version 3.5.1.
- Author network diagrams were generated in which authors of the same publications are connected by a straight line and sizes of nodes are proportional to the number of publications authored.

Results

Authorship

- A total of 285 publications were identified, with 1,394 experts listed as authors on ≥ 1 article.
- 282, 127 and 58 participated as authors on ≥ 2 , ≥ 3 and ≥ 4 publications, respectively (Figure 1).
- The maximum number of publications co-authored by an individual was 16.

First Authorship

- 32, 5 and 2 experts participated as first authors on ≥ 2 , ≥ 3 and ≥ 4 publications, respectively (Figure 2).
- The maximum number of first-author publications by an individual was 5 (Figure 2).

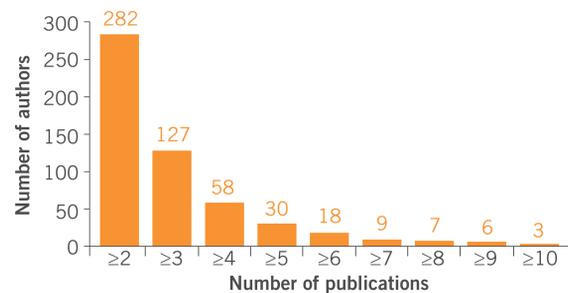
Author Networks

- Figure 3 shows an author network map detailing the collaborations of authors with ≥ 6 publications.
- Key contributors to the field of phenylketonuria appeared to be A MacDonald, FJ Van Spronsen and N Blau (Figure 3).

Conclusions

- The analyses reported here could provide essential information for researchers or publication professionals previously unfamiliar with the field of phenylketonuria and could be used in other diseases of interest.
- This tool enables researchers to determine the number of publications (first) authored by an expert, as well as their previous collaborations with other experts. This can help to elucidate key research areas as well as their most prominent contributors.
- This technique could be used by publication professionals in conjunction with publication planning tools such as gap analyses and literature reviews, to inform authorship decisions and expert opinion elicitation methods.

Figure 1 | Authors stratified by number of publications co-authored



1,394 experts, who had authored ≥ 1 publication, were identified.

Figure 2 | Authors stratified by number of first-author publications

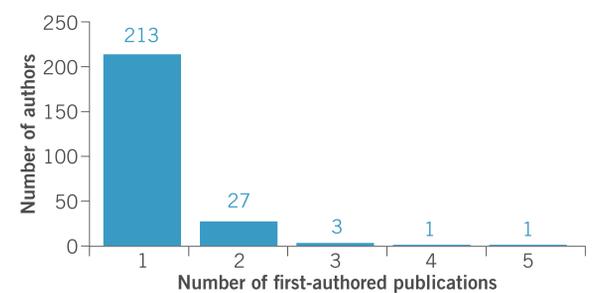
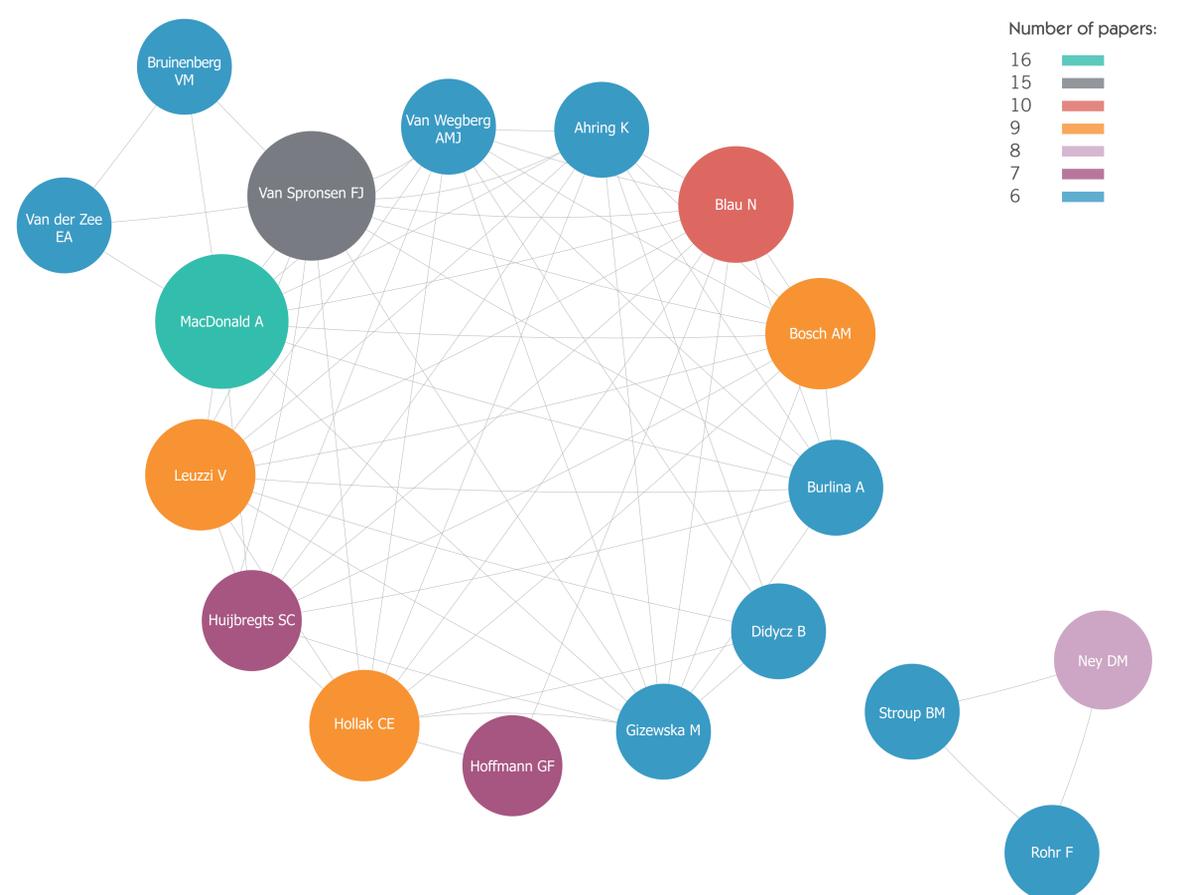


Figure 3 | Author network map showing authors of ≥ 6 publications



Node size is proportional to number of publications authored. The maximum node size indicates 16 publications authored.

Abstract

Objective

Identifying key opinion leaders (KOLs) can be difficult when looking to publish or establish guideline consensus, especially for researchers working in previously unfamiliar areas. This is particularly challenging in rare diseases, where there is often a need to identify a broad range of specialists to manage multi-system conditions, with only a limited number of disease-specific experts available. Using phenylketonuria as a case study, we aimed to identify KOLs by quantifying previous authorship contributions.

Research Design and Methods

Abstracts with the term "phenylketonuria" in their title or body text from 01/01/2016–19/09/2018 were identified via PubMed using a pragmatic literature review. Different variations of author names were assessed and compiled as required. Authors in the by line of ≥ 1 publication were selected and stratified according to number of phenylketonuria publications on which they had been listed as a (first) author using R version 3.5.1.

Results

285 publications were identified, and 1,400 experts were listed as authors on ≥ 1 articles. 282, 126 and 55 participated as authors on ≥ 2 , ≥ 3 and ≥ 4 publications, respectively. 7 experts co-authored ≥ 8 publications and the maximum number of publications co-authored by an individual was 16. 32, 5 and 2 had participated as first authors on ≥ 2 , ≥ 3 and ≥ 4 publications, respectively. The maximum number of publications first-authored by an individual was 5.

Conclusions

By elucidating the most prominent authors in phenylketonuria, these analyses provide essential information for researchers previously unfamiliar with this disease area. Publication professionals can use this technique alongside other publication planning tools such as gap analyses and literature reviews to rigorously support authorship decisions.

Author Contributions

Substantial contributions to study conception/design, or acquisition/analysis/interpretation of data: AH, ET, DS; Drafting of the publication, or revising it critically for important intellectual content: AH, ET, DS; Final approval of the publication: AH, ET, DS.

Acknowledgements

The authors thank Danielle Hart, Costello Medical, for graphic design assistance.