Reporting of Delphi Methods to Achieve Consensus on Guidelines in Rare Diseases

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Objective

- To analyse reporting of the Delphi method to achieve consensus on diagnosis and management guidelines in rare diseases.

Background

- The Delphi method is a structured process, widely used to achieve expert consensus during the development of guidelines for decision-making in clinical practice.[1]
- While the methodological quality of practice guidelines is often assessed using the Appraisal of Guidelines for Research and Evaluation (AGREE) Reporting Checklist,[2] there is currently a lack of standardised reporting requirements for Delphi methods. An e-Delphi checklist is currently in development.[3]
- Here, the use of the Delphi technique to achieve consensus on guidelines in rare diseases has been analysed.

Methods

- A pragmatic literature review was conducted in September 2017 by simultaneously searching Embase and MEDLINE® databases via the OvidSP platform.
- The search terms used related to rare diseases, practice guidelines and Delphi methodology **(Figure 1)**. No date limit was applied.

Figure 1 | Search terms

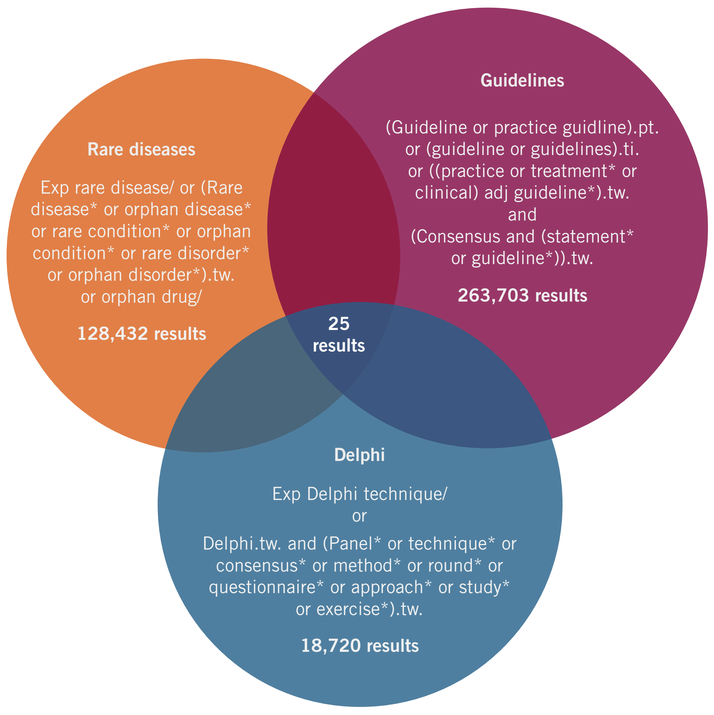


Figure 2 | PRISMA diagram

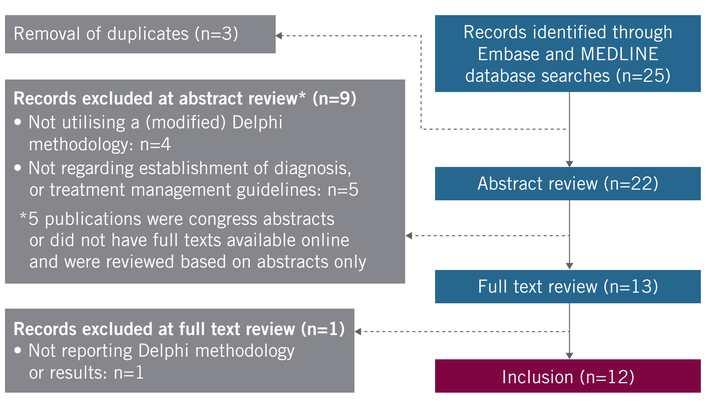
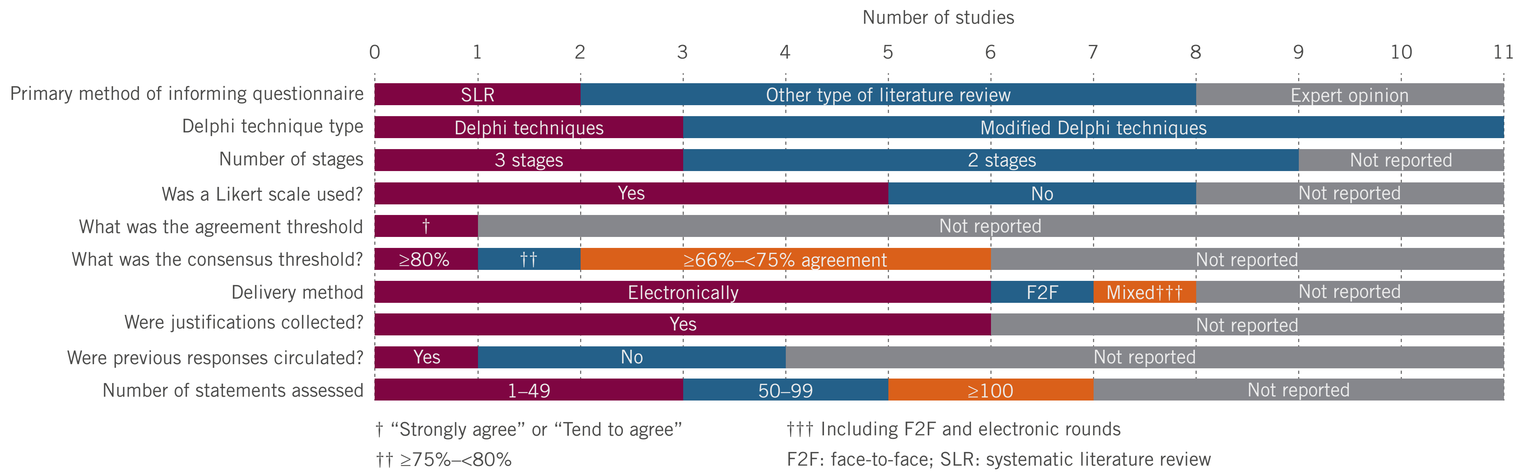


Table 1 | Summary of the rare diseases investigated within the identified publications

Condition	Number of studies	Type of guidelines	References
Orphanet-classified rare diseases			
C1 inhibitor deficiency	1	Management	Longhurst *et al.* (2015)[4]
Mucopolysaccharidosis type I	1	Management	De Ru *et al.* (2011)[5]
Osteopetrosis	1	Management and diagnosis	Wu *et al.* (2017)[6]
Paediatric acute recurrent pancreatitis and chronic pancreatitis	1	Diagnosis	Gariepy *et al.* (2017)[7]
Pulmonary arterial hypertension	1	Management	Rahaghi *et al.* (2016)[8]
Rett Syndrome	3	Management	Baikie *et al.* (2014)[9] Downs *et al.* (2009)[10] Leonard *et al.* (2013)[11]
Vasculitis	1	Management	Brown *et al.* (2013)[12]
Author-defined rare conditions			
Mesenteric ischaemia	1	Management	Oliva *et al.* (2013)[13]
Squamous cell carcinoma of the vulva	1	Management	Kidd *et al.* (2013)[14]

Figure 3 | Reporting of the Delphi method



† "Strongly agree" or "Tend to agree"

†† ≥75%–<80%

††† Including F2F and electronic rounds

F2F: face-to-face; SLR: systematic literature review

- Publications were screened by a single reviewer to include English language articles in which any version of the Delphi method was used for the development of guidelines for treatment, management or diagnosis of an Orphanet-classified rare disease or author-defined rare condition.
- Full texts of the eligible publications, and abstracts only where full texts were unavailable, were assessed for the quality of reporting.
- Results were evaluated against pre-specified criteria, which were based on the relevant section of the AGREE checklist.[2]

Results

Literature Review

- Searches identified 25 results, which were published between 2009–2017.
- 12 records fulfilled the inclusion criteria, which reported on 11 unique Delphi studies **(Figure 2)**.
- Within the 11 unique Delphi studies, 7 Orphanet-classified rare diseases and 2 author-defined rare conditions were investigated **(Table 1)**.
- All studies reported consensus results.

Reporting of the Delphi Method (Figure 3)

- Literature searches guided the development of statements for Delphi panel review in 8/11 (73%) studies, but only 2/11 (18%) conducted systematic literature reviews.
- The majority of studies (8/11, [73%]) used a modified Delphi method, while the remainder used a classic three-stage process.
- The rating scales used were not described by 3/11 (27%) studies and only 5/11 (45%) studies reported the use of Likert scales.
- Of greatest importance, agreement thresholds were not reported for 10/11 (91%) studies and the consensus threshold was not reported for 5/11 (45%) studies.
- The reported methods of delivering the results to the panelists were via email, face-to-face or a combination of the two. However, 3/11 (27%) studies did not report the method of delivery used.
- The collection and circulation of panelist feedback, which was used to inform subsequent Delphi stages, were not reported by 5/11 (45%) and 7/11 (64%) studies respectively.
- Only 7/11 (64%) reported the number of statements assessed.

Conclusions

- Since data from randomised controlled trials are scarce in rare diseases, well-designed Delphi studies present a valuable tool to establish consensus and develop relevant clinical guidelines.
- Improved reporting of Delphi methods in publications is necessary, with the majority of identified studies lacking AGREE-recommended detail.
- To improve reporting of Delphi-based consensus guidelines for rare diseases, publication should be carried out in line with existing and newly, developed best practice recommendations for reporting Delphi methods.

Abstract

Objective

- To analyse reporting of the Delphi method to achieve consensus on diagnosis and management guidelines in rare diseases.

Research Design and Methods

- A pragmatic literature review was conducted in September 2017 by searching EMBASE and MEDLINE® databases. Publications were screened by a single reviewer to include English language articles, in which the Delphi method was used for the development of guidelines in rare diseases. Reporting of the Delphi method was critiqued against the AGREE Reporting Checklist.

Results

- Searches identified 25 results published between 2009–2017, 12 of which fulfilled the inclusion criteria. 11 unique Delphi studies were identified, which investigated 7 Orphanet-classified rare diseases and 2 author-defined rare conditions; all studies reported consensus results. The majority (8/11 [73%]) used a two-stage modified Delphi method, while the remainder used a classic three-stage process. Literature searches guided the development of statements for Delphi panel review in the majority of studies, but only 2/11 (18%) conducted systematic literature reviews and merely 7/11 (64%) studies reported the number of statements assessed. Furthermore, 7/11 (64%) did not report collecting panellist feedback to inform subsequent Delphi stages, 5/11 (45%) studies did not describe the rating scales used, and 3/11 (27%) omitted reporting the level of consensus reached.

Conclusions

- There is a need for improved reporting of Delphi methods, with the majority of identified studies lacking AGREE-recommended detail. Since data from randomised controlled trials are scarce in rare diseases, well-designed Delphi studies are a valuable tool, and as such, publication should be considered and carried out in line with existing, and newly developed recommendations on best practice.

References

1. Boulkedid R. *et al.* PLoS One 2011;6(6):e20476; **2.** Brouwers M. *et al.* BMJ 2016;352:i1152; **3.** Guerreiro M. *et al.* Reporting E-Delphi Studies (REDS) in health research: development of a preferred items checklist 2016. Available at: www.equator-network.org/wp-content/uploads/2009/02/Protocol-E-Delphi-studies-reporting-guideline.pdf [Last accessed 22.11.17]; **4.** Longhurst H. *et al.* Clin Exp Immunol 2015;180(3):475–83; **5.** de Ru M. *et al.* Orphanet J Rare Dis 2011;6(1):55; **6.** Wu C. *et al.* J Clin Endocrinol Metab 2017;102(9):3111–23; **7.** Gariepy C. *et al.* J Pediatr Gastroenterol Nutr 2017;64(1):95–103; **8.** Rahaghi F. *et al.* Chest 2016;150(4 Suppl 1):1148A; **9.** Baike G. *et al.* J Pediatr Gastroenterol Nutr 2014;58(2):237–44; **10.** Downs J. *et al.* Spine 2009;34(17):E607–17; **11.** Leonard H. *et al.* J Pediatr Gastroenterol Nutr 2013;57(4):451–60; **12.** Brown N. *et al.* Presse Med 2013;42(4 Part 4): 746–7; **13.** Oliva I. *et al.* Abdom Imaging 2013;38(4):714–9; **14.** Kidd E. *et al.* Am J Clin Oncol 2013;36(4):415–22.

Author Contributions

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

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