Delphi hackathon – a new approach to develop Core Outcome Sets for blood cancers

Katharina Maria Lang

`katharina-maria.lang@charite.de`

Charité University Medicine Berlin https://orcid.org/0000-0001-8855-884X

Ellen de Waal
EHA: European Hematology Association

Tamás Bereczky
Patient Advocate Foundation

Dalia Dawoud
NICE: National Institute for Health and Care Excellence

Jan Geissler
Patient Advocate Foundation

Natacha Bolanos
: Leukemia and Lymphoma Society

Guillermo Sanz
Valencian Community Foundation Principe Felipe Research Center: Fundacion de la Comunidad Valenciana Centro de Investigacion Principe Felipe

Yann Guillevic
Bristol-Myers Squibb Company

Jesus Maria Hernandez Rivas
Universidad de Salamanca

Linda van den Berg
inScience Communications

Martje Barbus
AbbVie Deutschland GmbH & Co KG Ludwigshafen

Renate Schulze-Rath
Bayer Pharma AG

Lars Bullinger
Charite University Hospital Berlin: Charite Universitatsmedizin Berlin

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Research Article

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Abstract

Background

The Delphi technique is a well-known and accepted method to reach consensus. However, major challenges in Delphi studies include recruiting suitable participants and avoiding a loss of participants between survey rounds. To mitigate these challenges, we developed a virtual “Delphi hackathon” as a new method to conduct Delphi studies. We have used this new approach to develop Core Outcome Sets (COS) for hematologic malignancies within the scope of the HARMONY PLUS project - as part of the HARMONY Alliance,a public-private European Network established in 2017, which currently includes 53 partners and 76 associated members from 26 countries.

Methods

In the style of a classic hackathon, known from software development, we organized two virtual meetings where all participants simultaneously participated in two rounds of online Delphi surveys, followed by a third virtual meeting to reach final consensus. Recruitment of participants was done by advertising and providing information via the HARMONY Alliance communication team and patient umbrella organizations.

Discussion

Big challenges seen in previous Delphi surveys such as difficulties in the recruitment of the various stakeholders to participate in Delphi surveys and their continued support over consecutive Delphi rounds could be favourably addressed by our new Delphi hackathon approach, which showed superior results both with regard to recruitment and continuous support of participants.

Introduction

The Delphi technique is a method to reach consensus based on the results of surveys sent to a panel of experts (1). The Delphi method requires at least two rounds of a survey to reach consensus and a sustainable result between different stakeholder groups. In the first round of a Delphi survey, the participants are usually asked for their personal opinion. In the following round, this opinion is to be reconsidered based on the other participants' responses to the first round. For this purpose, the summarized results of all other participants are made available to all participants before they can revise their decision during the second round. Finally, the ultimate result is meant to constitute a consensus of the group’s opinion, which is usually obtained during a consensus meeting.

In medicine, the Delphi technique is used to develop Core Outcome Sets (COS) for diseases, i.e., the minimum set of outcomes that should be collected and reported in future clinical trials according to patients, clinicians, and other stakeholder groups. Developing COS for hematologic malignancies (HMs) is one of the objectives of the HARMONY Alliance. The HARMONY (Healthcare Alliance for Resourceful Medicine Offensive against Neoplasms in Haematology) Alliance is a public-private European Network established in 2017, which currently includes 53 partners, inter alia 6 cancer patient umbrella organisations, and 76 associated members from 26 countries. One of HARMONY Alliance's goal is to use Big Data to improve understanding and treatment of hematological malignancies. In order to achieve this aim, HARMONY PLUS (IMI 945406) is structured into six work packages of which Work Package 2 (WP 2) is focused on defining outcomes that are relevant to each hematological malignancy.

Within the framework of the HARMONY project (IMI 116026), surveys using the Delphi method have been conducted in recent years with the aim of defining COS for various malignant hematologic diseases (Fig. 1) (2).

Similar to other traditional Delphi studies (3), the HARMONY Delphi studies were run as electronic surveys that were open for a certain amount of time and were brought to the attention of potential participants through email or social media (Fig. 2). Many groups have described that the recruitment and adherence of participants between the individual Delphi rounds pose a great challenge (4). This was also confirmed by the experience from the Delphi surveys conducted by the HARMONY Alliance. In addition, the HARMONY Alliance had the challenge of achieving consensus among four stakeholder groups, i.e., patients, clinicians, drug developers, and regulators, which have different information needs and levels of scientific literacy, thereby supposing an additional challenge to conduct a stakeholder overarching COS definition using a Delphi survey.

Aims

Our experiences with traditional Delphi surveys revealed several challenges:

- very busy schedules of the participants;
- challenges in reaching suitable participants, partly due to the rare nature of some of the hematologic diseases concerned;
- different information needs and demands on the survey;
- limited participation of the same people in the second Delphi round.

Therefore, we have reconsidered and revised the way of conducting the surveys and developed a new approach: the Delphi hackathon.

Methods

The COS development will follow recommendations of the Core Outcome Measures in Effectiveness Trials (COMET) initiative from the international Core Outcome Set Standards for Development (COS-STAD) (1, 5).

A prospective study protocol was published on HARMONY webpage (2). The protocol has been written following the Core Outcome Set-Standardised Protocol (COS-STAP) recommendations in cooperation between all stakeholder groups (6).

The Delphi hackathon approach

In the style of a classic hackathon, we organized two virtual meetings where all participants simultaneously participated in two online Delphi surveys, followed by a third virtual meeting to reach final consensus (Figure 3). The first two meetings were scheduled at an interval of two weeks, lasted 90 minutes each, and covered four different hematologic diseases: Chronic Myeloid Leukemia (CML), Myeloproliferative Neoplasms (MPN), Hodgkin's Lymphoma (HL), and Waldenström Macroglobulinaemia (WM).

Recruitment mainly took place within the HARMONY community, targeting the four main HARMONY stakeholder groups - patients, clinicians, drug developers, i.e., members of the European Federation of Pharmaceutical Industries and Associations (EFPIA), and regulators. The recruitment was done by advertising and providing information via the HARMONY Alliance communication team and patient umbrella organizations. A protocol with detailed information on the study design and the planned analysis was developed and published before the hackathons took place (2). While some participants were invited to complete surveys for all four diseases, the majority (especially the patients) only completed the survey corresponding to their own expertise.

Delphi hackathon – study design

The first virtual meeting started with a brief plenary introduction to the methodology and the purpose of the survey presented by the study leaders, followed by a hands-on introduction with screenshots of the Delphi tool explaining how to carry out the self-registration and the survey in practice. After the plenary introduction, break-out rooms were made available for all participants in addition to the main plenum room. The questions of the participants could easily be answered by the study leaders in these break-out rooms. In addition to content-related questions, technical and organizational issues could also be discussed here. The Delphi surveys consisted of a list of approximately 60 outcomes for HMs, e.g., overall survival, progression free survival, cost of treatment, pain, and fatigue. The participants were asked to score the importance of each outcome on the list. After completion of the first round, the participants could leave the meeting.

In *the second meeting*, participants were asked to reconsider their initial ratings of the outcome measures in view of the results of the other participants. All participants of the first round received several email reminders of the date of the second meeting. Similar to the first meeting, a short introduction was given at the beginning of the second meeting, emphasizing the goal of the Delphi survey to reach consensus. Summarized results of the other participants were made available per stakeholder group. Again, participants could ask questions via chat at any time. At this second meeting, break-out rooms were not offered anymore, as all technical and organizational questions had already been addressed during the first meeting.

A *final consensus meeting* was planned after completion of the final analysis of the results of the second Delphi hackathon round. There was an eight-week interval between the second round and the final consensus meeting. This meeting also took place virtually to give everyone the opportunity to participate. The goal of this meeting was to confirm all the outcomes that had been ranked high in the COS, and to discuss those that had not met any of the pre-determined consensus criteria. The summary of the results was shared with all participants prior to the consensus meeting for transparency and to allow for better preparation and discussion.

Ethics approval and consent to participate

One of HARMONY PLUS' work packages is responsible for ethical aspects of the project. For comprehensive information see https://www.harmony-alliance.eu/work-packages-harmony-plus/work-package-6-legal-ethics-and-governance. In accordance, the Delphi hackathon was developed and conducted in alignment with HARMONY PLUS ethics approval. Recruitment of participants was made within HARMONY Alliance, and in addition patient participant recruitment was also made through patient umbrella organisations partnering with HARMONY. Before registration, all study participants received information about the nature and aims of study as well as their voluntary participation. Then, informed consent was obtained from all participants during the registration procedure prior to filling out the Delphi surveys.

Study management group

As recommended by the COMET initiative, a study management group was assembled to oversee the project (1). The group should comprise a study coordinator, a haematologist with a leading role in the treatment of HMs and the conduction of clinical trials in the field, a drug developer with experience in performing clinical trials, patient advocates and methodological experts with experiences of systematic reviews and Delphi studies. The role of the study management group was to support the development of the study protocol and to review the list of outcomes and the associated lay versions and descriptions.

Consensus criteria

To reduce potential bias in interpretation of the results a clear consensus definition is important. We used three categories of consensus that were already used in previous works (1, 3).

1. Consensus in = 70% or more over all respondents scored the outcome as critically important and 15% or fewer over all respondents scored the outcome as limited important
2. Consensus out = 70% or more over all respondents scored the outcome as limited important and 15% or fewer over all respondents scored the outcome as critical important
3. No Consensus

Outcomes that did not achieve consensus through two Delphi rounds were discussed in a face-to-face consensus meeting to finally ratify the core outcome sets. Representatives from all participating stakeholder groups were part of this meeting.

Analysis

Analysis of the Delphi study were using descriptive statistics. The results for each Delphi round, for each outcome and for each stakeholder group were presented in frequency tables. The analysis of the Delphi survey was performed using the R statistical software version 3.5.2.

As an exploratory analysis, we additionally identified outcomes considered as especially important for patients, irrespective of the other stakeholder groups. The median Likert score for the patient group at the end of each round was calculated and those outcomes achieving a median of greater or equal to 7 will be considered as important for patients and was included in the COS. In this way, patient-important outcomes were separately discussed in the final consensus meeting.

Discussion

Strengths and limitations

One of the strengths of the Delphi hackathon approach is that blocking dates in calendars helps people to dedicate their time and attention to the survey. Emails asking to answer a survey are easily overlooked in the flood of work emails. The dates for both rounds were announced in advance. In addition, personalized email reminders were sent out several times between the Delphi rounds. This was meant to avoid a loss of participants between rounds.

Of course, the fixed date can also be a disadvantage because some participants who are interested in the survey may not be available on the fixed dates. These votes and opinions would then be missed. To tackle this, the survey was also made available online for an additional ten days, together with explanatory information and recordings of the virtual meetings.

Another strength of our virtual Delphi hackathon approach is being able to respond to questions tailored to the information needs of individual participants. Experiences from the previous HARMONY surveys showed that the information needs varied greatly between the groups, and the hurdle to pose a question via email often was too high. Therefore, the possibility to immediately ask questions during the meeting – either in person or via the meeting chat was very convenient and heavily used. In addition, the introductory explanation of the background of the study motivated participants to complete the surveys, preventing them from dropping out.

Initially, there were concerns that the nature of the meeting might influence the outcome of the survey, because a major advantage of using the Delphi method is that all participants can cast their votes anonymously. However, by completing the survey in private and not via a split screen, etc. anonymity could be maintained.

The results of the Delphi hackathons (i.e., the Core Outcome Sets for the four HMs listed above) will be reported in a separate paper following a final expert panel meeting.

Trial status

At the time of manuscript submission, the Delphi hackathon was completed.

Abbreviations

CML – chronic myeloid leukemia

COS – core outcome set

COMET – Core Outcomes Measures in Effectiveness Trials

EFPIA – European Federation of Pharmaceutical Industries and Associations

HARMONY – Healthcare Alliance for Resourceful Medicine Offensive against Neoplasms in Hematology

HL – Hodgkin lymphoma

HMs – hematological malignancies

MPN – myeloproliferative neoplasms

WM – Waldenstroem’s Macroglobulinemia

WP - Workpackage

Declarations

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Availability of data and material

preliminary outcome lists for WM as an example

Consent for publication

Not applicable.

Competing interests

All authors are members of the HARMONY Alliance consortia, otherwise there are no conflicts of interest with regard to the Delphi study protocol. MB and RSR are employed by AbbVie and Bayer AG, respectively.

Authors' contribution

All authors made substantive contributions to the design of the study. KL, MB, RSR and TB elaborated the study protocol in detail; the co-authors critically reviewed it. KL drafted the manuscript, and all co-authors reviewed the manuscript and approved it to be published.

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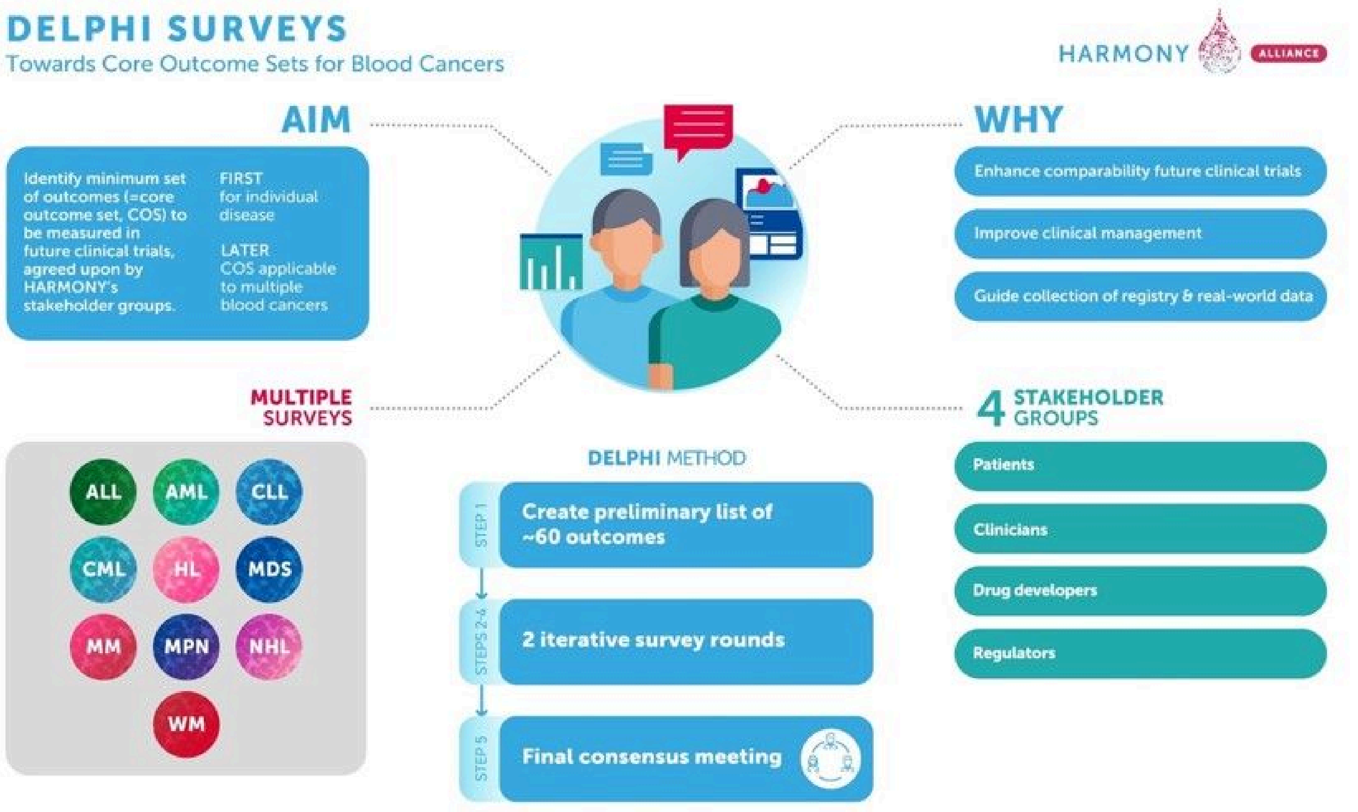
Figures



DELPHI SURVEYS

Towards Core Outcome Sets for Blood Cancers

HARMONY ALLIANCE

AIM

Identify minimum set of outcomes (=core outcome set, COS) to be measured in future clinical trials, agreed upon by HARMONY's stakeholder groups.

FIRST for individual disease

LATER COS applicable to multiple blood cancers

WHY

- Enhance comparability future clinical trials
- Improve clinical management
- Guide collection of registry & real-world data

MULTIPLE SURVEYS

ALL, AML, CLL, CML, HL, MDS, MM, MPN, NHL, WM

DELPHI METHOD

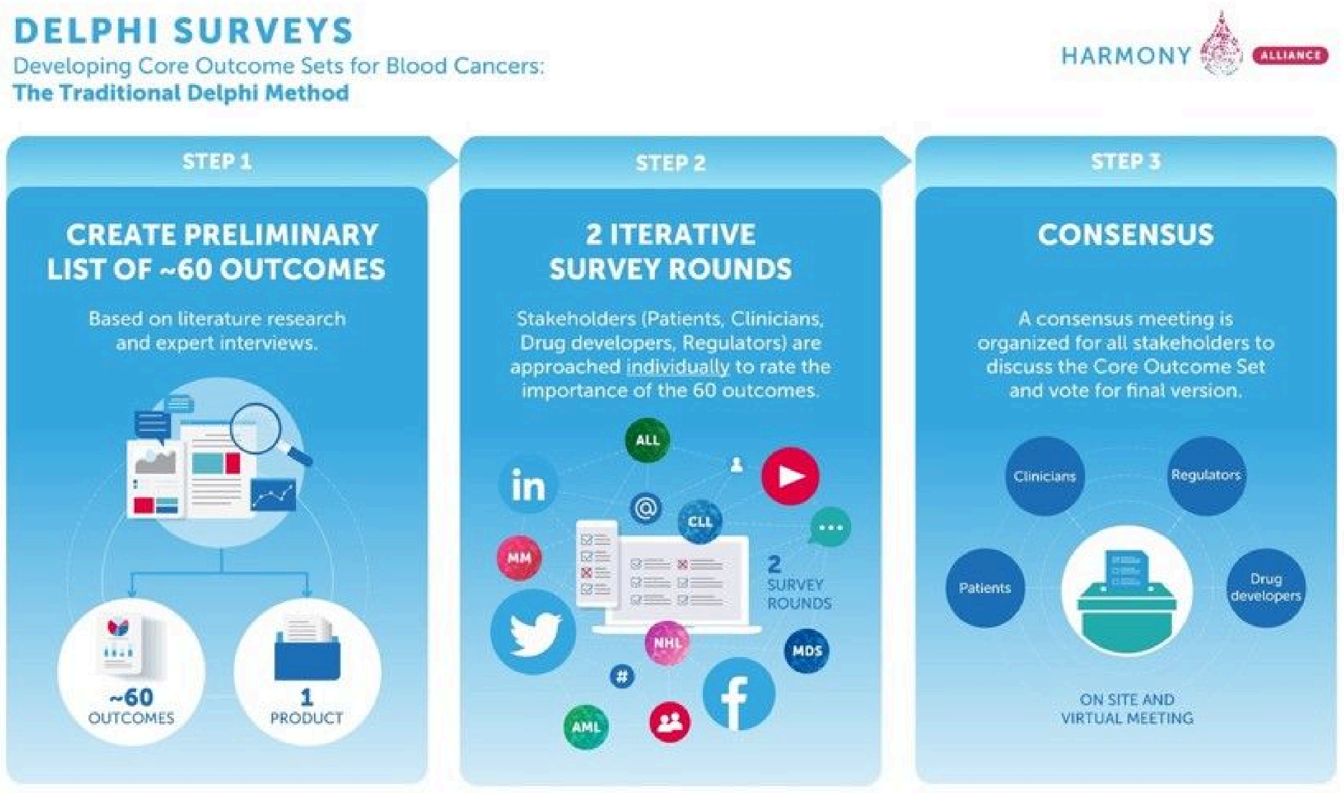
- STEP 1: Create preliminary list of ~60 outcomes
- STEPS 2-4: 2 iterative survey rounds
- STEP 5: Final consensus meeting

4 STAKEHOLDER GROUPS

- Patients
- Clinicians
- Drug developers
- Regulators

Figure 1

Infographic of the HARMONY Alliance Delphi Survey method



DELPHI SURVEYS

Developing Core Outcome Sets for Blood Cancers:

The Traditional Delphi Method

HARMONY ALLIANCE

STEP 1

CREATE PRELIMINARY LIST OF ~60 OUTCOMES

Based on literature research and expert interviews.

~60 OUTCOMES

1 PRODUCT

STEP 2

2 ITERATIVE SURVEY ROUNDS

Stakeholders (Patients, Clinicians, Drug developers, Regulators) are approached individually to rate the importance of the 60 outcomes.

STEP 3

CONSENSUS

A consensus meeting is organized for all stakeholders to discuss the Core Outcome Set and vote for final version.

ON SITE AND VIRTUAL MEETING

Figure 2

HARMONY Alliance Delphi Survey - traditional method


DELPHI SURVEYS
Developing Core Outcome Sets for blood cancers:
The Delphi Hackathon

STEP 1
CREATE PRELIMINARY LIST OF ~60 OUTCOMES
Based on literature research and expert interviews.

STEP 2
2 REAL TIME SURVEY ROUNDS
Interactive meetings are organized to invite stakeholders (Patients, Clinicians, Drug developers, Regulators) to discuss and rate real-time the importance of the 60 outcomes.

STEP 3
CONSENSUS
A consensus meeting is organized for all stakeholders to discuss the Core Outcome Set and vote for final version.

Figure 3

HARMONY Alliance Delphi Survey – hackathon method

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

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