



Improvements and advances in core outcome set methodology: proceedings of the CS-COUSIN & COMFA Joint Meeting

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Abstract

A COS is a systematically developed list of outcomes recommended for reporting all studies within a particular disease area. The Cochrane Skin–Core Outcome Set Initiative (CS-COUSIN) and the Core Outcome Measures in Food Allergy (COMFA) Initiative Joint Meeting was held virtually on September 23 through 28, 2021. This meeting brought together core outcome set (COS) developers within the domains of dermatology and food allergy, respectively. The meeting assessed the state of COS methodology, provided a forward look, and facilitated the networking of members of individual COS groups. The meeting hosted several expert-led key addresses and focus sessions, in which work was critically discussed, new developments were shared, and groundwork was laid for future projects. The conference emphasized the importance of ongoing COS work to improve patient outcomes and limit research waste.

Keywords Core outcome set · Methodology · Development · Proceeding

Introduction

The methodology for core outcome set development has continued to evolve over the past decade. Work has been done to produce core outcome sets that are inclusive and generalizable, and ensure the development process minimizes the introduction of the specific biases of the various stakeholders [2, 10, 13, 15]. In particular, the patient perspective is recognized as extremely important. Further advances in core outcome set methodology were discussed at the recent

Cochrane Skin–Core Outcome Set Initiative (CS-COUSIN) and Core Outcome Measures in Food Allergy (COMFA) Initiative Joint Meeting, held virtually September 23 through 28, 2021 in Magdeburg, Germany (<https://cconline.eventsair.com/cs-cousin/>) [9]. Both CS-COUSIN and COMFA are dedicated to the development of core outcome sets (COSs), which are standardized minimum outcomes for reporting findings from clinical trials and other studies within a given disease area [14, 18]. CS-COUSIN is an umbrella organization for COS groups which develop dermatological outcome

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sets, while the COMFA Initiative develops core outcomes related to food allergies [7, 9, 14]. COMFA is a COST Action (CA18227), which receives funding supported by COST (European Cooperation in Science and Technology, www.cost.eu) [8]. The conference was jointly chaired by Professor Christian Apfelbacher (CS-COUSIN, co-chair of HOME and HECOS and grand holder scientific representative of COMFA) and Dr. Daniel Munblit (COMFA chair).

The overall aim of this conference was to apprise researchers of the state of COS development and to provide a space for researchers in the field of COS methodology to learn from the experiences of others [9]. Attendees were afforded opportunities for collaboration and to progress their own work through dedicated focus sessions [9]. Additionally, COMFA hosted a training school, which aimed to provide supplementary guidance and support for early researchers [9].

Core outcome sets are developed to guide future studies within a given disease area to consider the most important factors as rated by physicians, patients, and other stakeholders alike [14, 18]. The goal of COS work is to increase consistency of outcome reporting within a disease area, as well as provide a unified consideration of the most important outcomes as perceived by stakeholders [18]. In doing so, there will be uniform trial outcome analysis, as well as ensured consistency between reported results [18]. The Core Outcome Measures in Effectiveness Trials (COMET) Initiative, a leading initiative in the field of COS, as well as CS-COUSIN, have established informational guidelines and helpful material for standardized stages in COS development that all project groups are encouraged to use [6, 14]. Additionally, the HOME (Harmonizing Outcome Measures for Eczema) roadmap provides dermatological COS projects a useful outline on which to build a sound outcome set [1, 16]. This communication provides a brief summary of the high-quality work that was done within the working groups, as well as the collective experience of the conference attendees.

Keynote addresses

The Joint Meeting was formally initiated by a keynote address entitled ‘*Lessons Learned from COS Development in Long COVID*’ by Dr. Daniel Munblit and Professor Paula Williamson. Dr. Munblit is affiliated with the Institute of Child’s Health at Sechenov First Moscow State Medical University, the National Heart and Lung Institute of the Imperial College London, and the Research and Clinical Center for Neuropsychiatry in Moscow, Russia. Professor Williamson is affiliated with the University of Liverpool Biostatistics Department. The opening address provided an overview of core outcome sets, including the COMET 2010 definition, the usefulness of COSs, and previous COVID COS project lessons learned,

including a study by Tong et al. entitled ‘*Core Outcomes Set for Trials in People with Coronavirus Disease 2019*’ [17]. This was followed by an in-depth discussion of the ongoing development of a core outcome set for Long COVID, details of which can be found on the COMET database [11]. Following, Patient Partner Maureen Smith gave a keynote entitled ‘*Involving Patients in Core Outcome Set Development: Reflections From a Patient Partner*.’ Ms. Smith serves on the Canadian Organization for Rare Disorders (CORD) Board of Directors, among other roles. She addressed the need for patient involvement in research and detailed the importance of including the patient perspective. The first day closed with an address given by Prof. Dr. med. Jochen Schmitt (Medical Faculty in the Department of Dermatology at the Technical University of Dresden) and Prof. Dr. Jan Kottner (Department of Dermatology and Allergy at the Charité University of Medicine in Berlin; Faculty of Medicine and Health Sciences at Ghent University) entitled ‘*CS-COUSIN—Where have we come so far?*’ CS-COUSIN’s presence within dermatology core outcome set development was detailed, as well as the goals, structure, leadership, and project group membership, details of which can be found online at <http://cs-cousin.org> [5]. Additionally, challenges including slow progress of output and a careful reflection on these trials were provided. Finally, the plenary session closed with a special announcement of the merger between CS-COUSIN and CHORD (Consortium for Harmonizing Outcomes Research in Dermatology; website accessed via <https://www.chordoutcomes.org/> [3]) into the CHORD-COUSIN Collaboration (C3). C3 is an exciting initiative to streamline dermatologic core outcome development under a unified umbrella organization. Finally, the closing day included a keynote address from Dr. David Cella, professor and Chair of the Department of Medical Social Sciences at Northwestern University, Feinberg School of Medicine and Director of the Institute for Public Health and Medicine—Center for Patient-Centered Outcomes. Entitled ‘*The relevance of PROMIS to the development of the Core Outcome Measures*,’ the address described PROMIS (Patient-Reported Outcomes Measurement Information System) and the role it can play in the future of COS (<https://www.healthmeasures.net/explore-measurement-systems/promis>) [12]. With the diverse topics covered in the plenary sessions for all attendees, there was ample opportunity for groups to glean generalizable information that can provide groundwork to facilitate discussion and momentum, as well as directive advice for those who are just beginning the process of such an undertaking.

Groups in attendance

Project groups were allotted focus session time to further the progress of their work through one of three key objectives, which included: reaching consensus on domains/

subdomains/outcomes, or agreement on a given problem; discussion of major achievements or progress that has been made; identification of next steps and best approaches [9]. In addition, each group was invited to describe this progress at the final day plenary session as a way to demonstrate key ideas which may prove useful to other participants. Groups in attendance and who provided a summary presentation included COSCAM, CORALS, HECOS, COMFA, HOME, IMPROVED, and UPGRADE, each of which is described in detail in Table 1.

Working group key takeaways

To summarize the high-quality work that was done over the course of the conference, each working group was invited to provide a presentation regarding the activities and key takeaways of their focus session. These provided insight into challenges, strategies, and successes, facilitated communication, and created a community of shared knowledge among groups. The first speaker, Dr. Ginger Beau Langbroek from the Department of Surgery at Amsterdam University Medical Centers, provided a high-level overview of COSCAM (Core Outcome Set for CApillary Malformations (COSCAM) [4]. She described COSCAM's methods and results, with detailed results of participants, outcomes for voting, and the final successful development of a core domain set for capillary malformations. Dr. Rosalind Simpson, associate professor at the Centre of Evidence-Based Dermatology at the University of Nottingham School of

Medicine, spoke on behalf of CORALS (Core outcomes for research in lichen sclerosus) [4]. She described where the group stands in the development phase, as well as the fruitful discussion held during the focus session. This included discussion surrounding the need to increase diversity and expand across more international areas, as well as the need to include adverse events in outcome sets. CORALS also noted the importance of maintaining momentum, which applies to all other project developers. Dipl.-Soz. Henriette Rönsch, Department of Dermatology of the University Hospital Carl Gustav Carus Technical University of Dresden, presented on behalf of HECOS (Development of a core outcome set for hand eczema) and gave a detailed overview of the progress that was made during the course of the conference [4]. Firstly, HECOS ratified and discussed the long list of outcomes that will be used in the Delphi Process, which consists of multiple rounds of surveys which collect ratings of importance for every outcome to ultimately determine the final list of outcomes following a final consensus meeting and vote. She also detailed thorough discussion of definitions, and the next steps of this project, which include translating the list into more than one language and increasing the number of patient participants. On behalf of COMFA, Dr. Daniel Munblit gave an overview of the systematic review that was performed, as well as a detailed description of the ongoing projects, with a conference output of finalized protocol development. In addition, the COMFA training school was a highly productive introduction to COS methodology for budding researchers or professionals that are new to the field of core outcome methodology, and covered systematic reviews, foundations of COS development, and

Table 1 Description of COS project groups in attendance

Group name	Primary condition(s)	Group lead(s)
Core Outcome Set for CApillary Malformations (COSCAM) [4]	Capillary malformations (port wine stains)	Ginger Beau Langbroek
Core outcomes for research in lichen sclerosus (CORALS) [4]	Genital lichen sclerosus	Rosalind Simpson
Development of a core outcome set for hand eczema (HECOS) [4]	Hand eczema	Christian Apfelbacher Andrea Bauer
The Core Outcome Measures for Food Allergy (COMFA) [7]	Food allergy	Christian Apfelbacher Daniel Munblit Robert Boyle
Harmonising Outcome Measures for Eczema (HOME) [1]	Atopic eczema	Eric Simpson Christian Apfelbacher
Measurement of Priority Outcome Variables in Dermatologic Surgery (IMPROVED) [4]	Facial aging; basal cell carcinoma; cutaneous squamous cell carcinoma; melasma; post-inflammatory hyperpigmentation; rosacea	Murad Alam
Understanding Pyoderma Gangrenosum: Review and Analysis of Disease Effects (UPGRADE) [4]	Pyoderma Gangrenosum	Alex G Ortega-Loayza

*Information from [1, 4, 7]

effective management of a COS development project. Prof. Kim Thomas, Centre of Evidence-Based Dermatology of the Queen's Medical Centre, University of Nottingham Faculty of Medicine and Health Sciences, and a leading member of HOME, reviewed the goals of the conference focus session, specifically the importance of “supporting implementation of the COS,” which HOME has successfully developed. As a collective group, they implemented pre-meeting surveys to identify opportunities or challenges in application and use of the COS. They subsequently held small-group discussions regarding the most important areas of focus, after which everyone had the opportunity to present an idea on how to overcome or address this challenge. Next, Bianca Younseon Kang, medical student at Creighton University School of Medicine, presented on behalf of the IMPROVED Group (Measurement of Priority Outcome Variables in Dermatologic Surgery) [4]. Ms. Kang presented a summary of the pre-Delphi kickoff workshop held with international physicians and researchers to present key ideas regarding conduct of an international COS and facilitation of a virtual consensus meeting. At the workshop, the results of a systematic review were evaluated, and attendees provided input regarding the most important questions to ask, the importance of involving patients and other important stakeholders in a similar workshop, and the need to include all specialties in the study. Finally, Dr. Alex G. Ortega-Loayza, Oregon Health and Science University School of Medicine associate professor of Dermatology, presented the UPGRADE (Understanding Pyoderma Gangrenosum: Review and Analysis of Disease Effects) working group discussion, which included “generation of items” for the long list of outcomes, as well as “domain classification”, i.e., which outcomes would fall into which domain category for Delphis [4]. The conference was an opportunity for UPGRADE to identify challenges and receive feedback, specifically on expanding and diversifying expert and patient involvement.

Conclusion

The CS-COUSIN & COMFA Joint Meeting was an opportunity for research professionals, physicians, and patients to come together to advance the development of core outcome sets and improve the associated methodology. The conference allowed individual working groups an opportunity to address important challenges and next steps. Individual COS groups also shared the status of their work with the larger group of conference participants. The merger of CS-COUSIN and CHORD into a unified organization, C3, was announced as an important step toward providing dermatology COS developers a cohesive parent organization. Key take-home messages included the importance of sound methodological preparation early in the COS development

planning stages; the need to include all stakeholder groups, including patients, in every phase of the process; and the importance of plans for uptake of COS work once produced.

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Declarations

Conflict of interest Murad Alam is Editor-in-Chief of Archives of Dermatological Research. McKenzie A. Dirr has no conflicts of interest. Karl Philipp Drewitz has no conflicts of interest. Nikita Nekliudov and Nina Seylanova have no conflicts of interest. Christian Apfelbacher is Section Editor for Outcomes Research of BJD and co-chair of the HOME and HECOS initiatives. Further, he received institutional funding from the Dr Wolff Group, and consultancy fees from the Dr Wolff Group, Sanofi Genzyme, Sanofi-Aventis Deutschland and LeoPharma.

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