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Establishing core domain sets for Chronic Nonbacterial Osteomyelitis (CNO) and Synovitis, Acne, Pustulosis, Hyperostosis, Osteitis (SAPHO): A report from the OMERACT 2020 special interest group

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ABSTRACT

Objective: A working group was established to develop a core domain set (CDS) for Chronic Nonbacterial Osteomyelitis (CNO) and Synovitis, Acne, Pustulosis, Hyperostosis, Osteitis (SAPHO) following the OMERACT filter 2.1.

Abbreviations: CNO, Chronic nonbacterial osteomyelitis; CDS, Core Domain Set; OMERACT, Outcome Measures in Rheumatology; SIG, Special Interest Group; SAPHO, Synovitis, Acne, Pustulosis, Hyperostosis, Osteitis

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Pustulosis
Hyperostosis
Osteitis
OMERACT
Outcome research

Methods: A scoping review to identify disease-related manifestations was performed, followed by a special interest group (SIG) session at OMERACT2020 to begin the CNO/SAPHO CDS framework.
Results: Candidate items were identified from the scoping review and most fell under Life Impact and Pathophysiology Manifestation core areas. A SIG agreed on the need to develop a CDS for CNO and SAPHO (100%) and for children and adults (91%).
Conclusion: Based on candidate items identified, qualitative research and Delphi surveys will be performed as next steps.

Introduction

Chronic nonbacterial osteomyelitis (CNO) is an autoinflammatory bone disease of unknown cause. While some patients develop unifocal and time-limited disease, most experience chronically active or recurrent disease affecting multiple bones, known as chronic recurrent multifocal osteomyelitis (CRMO) [1]. CNO is most frequently diagnosed in children and adolescents but can extend into adulthood [2,3]. A similar disease Synovitis, Acne, Pustulosis, Hyperostosis, Osteitis (SAPHO) primarily presents in adults, but can also manifest in children and adolescents [4]. SAPHO may be part of CNO's disease spectrum or its presentation in a different age group [5]. CNO commonly presents with bone pain, normal or mildly elevated inflammatory markers and radiographic abnormalities. Complications can include bone deformities, disfigurement, vertebral compression fractures, and leg length discrepancy.

Unfortunately, no known clinical or laboratory markers reliably measure disease activity in response to treatment. Some response criteria sets have been developed for CNO and SAPHO: the PedsCNO score, the CNO consensus treatment plan criteria for treatment failure, and a SAPHO osteitis and skin score [6,7,8]. However, none of these measures include input from patient and caregiver stakeholders on which items constitute the disease and are of major importance to them.

The aim of the OMERACT CNO/SAPHO working group is to develop a core domain set (CDS) for CNO and SAPHO that fulfills the requirements of the OMERACT 2.1 filter and can ultimately be used in clinical trials and observational studies [9,10,11]. Our goals at this initial stage were: 1) to perform a scoping review to identify existing disease-related manifestations and disease impacts previously reported for patients with CNO and SAPHO and 2) to obtain a consensus from physicians, researchers, patients and other stakeholders on a research agenda.

Materials and methods

Literature review

An initial search was conducted for existing CDS recommendations for CNO and SAPHO via the COMET data source (COMET database. http://www.comet-initiative.org/studies/search). A scoping review was performed in accordance with the recommendations of the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) [12,13]. Articles were searched electronically in OVID and PubMed databases on October 24th, 2019. The search protocol is available in Appendix 1. Selected PICOC (Population, Intervention, Control, Outcome, Context) criteria included patients of any age with CNO/SAPHO and any intervention, with 'standard of care' as the control. We included all observational study designs and case series with 3 or more patients to identify all available literature for CNO and SAPHO, given that these are rare diseases. Languages other than English were excluded.

Title and abstract screening, full-text screening, and article data extraction were performed by a group of practitioners and researchers (MO, YZ, CA, FN, MR, MH, GS, EF, AJ, SS, AL, AA, EW). Full-text manuscripts were obtained for all titles that fit the eligibility criteria. Independent reviewer pairs performed screening; any conflicts were resolved by a third reviewer. Extracted data included first author, study publication year, aim of the study, study design, population characteristics, all reported disease-related manifestations and impacts and their definition, any outcome measurement instrument used, and adverse events. All steps of the article selection process were documented using Covidence online platform (https://www.covidence.org/)

OMERACT 2020 SIG

A Special Interest Group (SIG) session was held for the CNO and SAPHO working group at the 2020 OMERACT virtual meeting on October 23rd, 2020. Participants were recruited through an established CNO research workgroup and through solicitation of other OMERACT members from the OMERACT organizers. The purpose of the SIG was to establish the need for a CDS for CNO/SAPHO and determine the feasibility of including both disease entities in this work, as well as adult and pediatric populations within a single CDS.

Results

Literature review

The scoping review yielded 4821 articles (Fig. 1). Following the removal of duplicates and abstract screening, 583 full-text studies were reviewed. Of these, 310 were excluded due to ineligible study type, patient population, or non-English language. The remaining 273 articles were eligible for data charting. The search did not yield any randomized controlled trials for CNO or SAPHO. Among 161 observational studies, there were 121 retrospective, 22 prospective, 11 case-control, and 7 cross-sectional studies. Case series evaluating 3 or more patients were included; there was a total of 97 case series with 13 having more than 10 patients and 84 with fewer than 10 patients. Three diagnostic and 12 basic science research studies were included. At the time of the 2020 OMERACT CNO SIG session, 121 articles were extracted by a primary reviewer and 25 were verified by a secondary reviewer.

Table 1 displays the preliminary disease-related manifestations and impacts reported from the scoping review mapped to core areas. The majority of items have been categorized under the Life Impact and Pathophysiology Manifestation core areas. Life Impact core area examples include quality of life, function, and pain. Pathophysiology Manifestations core area items include radiographic changes, inflammatory markers, and physical exam findings, such as limb swelling.

OMERACT 2020 SIG

There was a total of 28 participants at the OMERACT 2020 SIG session which included 26 clinicians and researchers, 6 of whom were adult rheumatologists, and 2 patient research partners.

The group first discussed limitations with prior outcome measures reported in the literature and the rationale for developing new outcome criteria and measure sets for CNO and SAPHO. An important concern is that prior published measures did not include the perspectives of patients and parents. Participants agreed that a core domain set must be developed that is relevant to all stakeholders and includes patient viewpoints. Another challenge was that certain items in previous outcome measures could not universally be used for all patients with CNO and SAPHO. Lastly, since many physicians use imaging to assess disease activity, there is a need for a validated

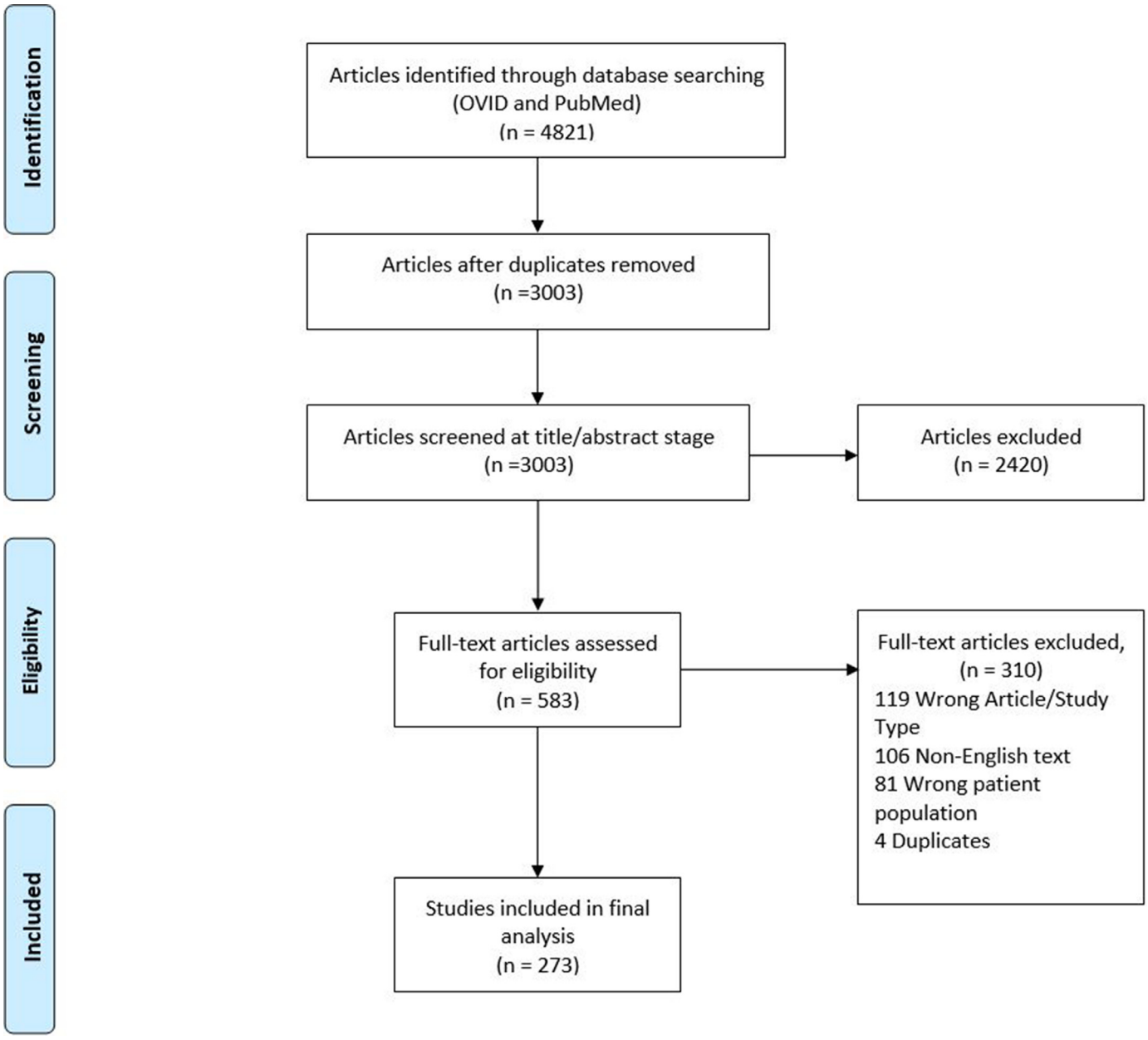

Fig. 1. PRISMA flow diagram for the scoping review.

quantifiable measurement of imaging. In the end, all participants voted and unanimously agreed (100%) on the need for a CDS for both CNO and SAPHO to be developed using OMERACT methodology.

The next discussion item focused on whether there should be separate or one all-inclusive CDS for CNO and SAPHO. The reasons in favor of separating the CDS included growth concerns in children

Table 1
Preliminary reported disease metrics mapped to core areas.

Core Areas Life impact	Pathophysiological manifestation	Resource Use/Economical impact	Death/Longevity
▪ Health Related Quality of life ▪ Overall health ▪ Physical activity ▪ Extent of skin involvement ▪ Pain ▪ Morning stiffness ▪ Functional impairment of extremities ▪ Asymmetric appearances of extremities/thorax ▪ Severity of disease per physician ▪ Severity of disease per patient	▪ Bone enhancement on imaging ▪ Change/Persistence of Radiograph findings ▪ Development of new lesions ▪ Local bone/tissue swelling ▪ Intestinal inflammation ▪ Inflammatory markers ▪ Sclerosis ▪ Change in skin findings ▪ Reduction in swelling ▪ Neutrophil activity ▪ Pro-inflammatory and the anti-inflammatory cytokines		

which do not apply to adults, different functional and quality-of-life concerns in children versus adults, and differences in organ system involvement between children and adults (e.g., adults are more likely to have skin involvement). The main reason in favor of an "all-inclusive" CDS is the clinical overlap between CNO and SAPHO. The previously published diagnostic criteria of SAPHO includes osteomyelitis and the only subset of SAPHO patients who do not meet CNO diagnostic criteria are those without bony but articular inflammation associated with skin conditions. [14] A suggested approach was to develop a single core set with separate mandatory domains for specific circumstances or optional domains for CNO and SAPHO. After discussion, the group voted on whether to combine or separate the CDS; 48% voted for CNO and SAPHO to have one core domain set, 43% voted for CNO to be the mandatory domain set with SAPHO mandatory in certain circumstances and 9% voted to separate the CDS.

Finally, the group discussed whether the CDS should cover both pediatric and adult populations, as outcomes of interest may differ between age groups. It was pointed out that at this stage of CDS development, the focus is on the domains that represent a disease rather than specific outcome measures and instruments. It was noted that if the underlying disease process is the same with an evolution from childhood to adulthood, then the domains could be the same with different instruments used based on age. The group voted and 91% were in favor of combining both pediatric and adult populations.

Discussion

The lack of widely accepted response criteria that include validated outcome measures and input of patients and parents emphasizes the need for the OMERACT CNO/SAPHO working group to develop a CDS for CNO and SAPHO. This was unanimously agreed upon at the recent OMERACT 2020 SIG session.

From the working group's initial scoping review, several candidate core set items used in previously published research studies were identified. Future work of the group will be to categorize the remaining disease-related items reported from the articles and refine the aggregate lists through nominal group exercises into the OMERACT core areas [11]. Finalized candidate items will form the basis of a proposed CDS for use in future clinical trials.

A significant majority of the voting group recommended combining CNO and SAPHO into one CDS rather than two separate CDS. An alternate approach is to have a mandatory CNO domain set to capture issues related to bony inflammation (common to both CNO and SAPHO) with additional SAPHO domains mandatory in specific circumstances. As the group continues to move through the OMERACT process and begins the qualitative research portion, further discussion regarding the organization of the CDS is planned.

Future research agenda

After the OMERACT CNO/SAPHO Working Group completes the scoping review, the group will next refine the list of potential items through nominal group exercise. In parallel, online focus groups will be conducted with patient and caregiver stakeholder groups to gain insight into what they deem essential for assessing disease domains. The group will also engage with additional adult rheumatologists, patients, caregivers and regulatory and industry representatives in this process to better represent all stakeholders' perspectives when developing the CNO/SAPHO CDS.

Author statement

Melissa Oliver: Writing- Original Draft, Writing- Review & Editing, Conceptualization, methodology, visualization, investigation. **Aruni Jayatilleke**: Writing- Review & Editing, Investigation, Methodology. **Eveline Wu**: Writing- Review & Editing, Investigation, Methodology. **Farzana Nuruzzaman**: Writing- Review & Editing, Investigation, Methodology. **Cassyanne Aguiar**: Writing- Review & Editing, Investigation, Methodology. **Aleksander Lenert**: Writing- Review & Editing, Investigation, Methodology. **Micol Dr.Romano**: Writing- Review & Editing, Investigation, Methodology. **Gabriele Simonini**: Writing- Review & Editing, Investigation, Methodology. **Samir Shah**: Writing- Review & Editing, Investigation, Methodology. **Matthew Hollander**: Investigation, Methodology, Writing- Review & Editing. **Alhanouf Alsaleem**: Investigation, Methodology, Writing- Review & Editing. **Emily Fox**: Investigation, Methodology, Writing- Review & Editing. **Alex Theos**: Methodology, Writing- Review & Editing. **Athimalaipet V Ramanan**: Writing- Review & Editing, Methodology. **Anja Schnabel**: Methodology, Writing- Review & Editing. **Sivia Lapidus**: Methodology, Writing- Review & Editing. **Lori Tucker**: Writing- Review & Editing, Methodology. **Karen Onel**: Methodology, Writing- Review & Editing. **Seza Ozen**: Writing- Review & Editing, Methodology. **Fatma Dedeoglu**: Writing- Review & Editing, Methodology. **Polly Ferguson**: Methodology, Writing- Review & Editing. **Beverley Shea**: Writing- Review & Editing, Resources, Supervision, Visualization, Methodology. **Jonathan Akikusa**: Writing- Review & Editing, Resources, Supervision, Methodology. **Christian Hedrich**: Writing- Review & Editing, Resources, Supervision, Methodology. **Philip Mease**: Writing- Review & Editing, Resources, Supervision, Visualization, Methodology. **Yongdong Zhao**: Writing- Review & Editing, Conceptualization, Methodology, Resources, Investigation.

Declaration of Competing Interest

None related to this work.

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Appendix 1

Search strategies:

- Databases searched: OVID, PubMed
- Dates of last search: OVID (10/24/19), PubMed (10/24/19)
- Search strategies:
  - OVID Search Criteria
    - Database: Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Daily and Versions(R) ⟨1946 to July 18, 2019⟩
    - Search Strategy:
    - 1. (Chronic adj (nonbacterial or recurrent multifocal) adj3 osteomyelitis).ti,ab.
    - 2. ((CNO or CRMO or clavicular or mandibular or Garre's or sternoclavicular) and (osteitis or osteomyelitis)).ti,ab.
    - 3. Majeed syndrome.ti,ab.
    - 4. chronic recurrent multifocal osteomyelitis.ti,ab.
    - 5. Chronic sclerosing osteitis.ti,ab.
    - 6. Diffuse sclerosing osteomyelitis.ti,ab.

- 7. autoinflammatory bone disorder.ti,ab.
- 8. SAPHO.mp. and osteitis.ti,ab. [mp=title, abstract, original title, name of substance word, subject heading word, floating subheading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
- 9. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8
- 10. (exp animal/ or exp invertebrate/ or animal experiment/ or animal model/ or exp plant/ or exp fungus/) not exp human/
- 11. 9 not 10
- 12. exp "Outcome Assessment (Health Care)"/
- 13. (outcome adj (domain* or assessment* or measure*)).ti,ab.
- 14. or/12–13
- 15. 9 and 14
- 16. (Subacute adj7 chronic adj7 osteomyelitis).ti,ab.
- 17. 16 or 11

- PubMed Search Strategy
  - PubMed Medical Subject Headings (MeSH) terms:
  - ((((((((((((((((((((((("bone and bones"[MeSH Terms] OR ("bone"[All Fields] AND "bones"[*All Fields*]) OR "bone and bones"[All Fields] OR "bone"[*All Fields*]) AND lesions[All Fields] AND ("acne vulgaris"[MeSH Terms] OR ("acne"[All Fields] AND "vulgaris"[*All Fields*]) OR "acne vulgaris"[All Fields] OR "acne"[*All Fields*]) AND fulminans[*All Fields*]) OR (Chronic[All Fields] AND multifocal[All Fields] AND ("osteomyelitis"[MeSH Terms] OR "osteomyelitis"[*All Fields*])) OR "Chronic recurrent multifocal osteomyelitis"[*All Fields*]) OR "Chronic sclerosing osteitis"[*All Fields*]) OR (Chronic[All Fields] AND symmetric[All Fields] AND ("osteomyelitis"[MeSH Terms] OR "osteomyelitis"[*All Fields*]))) OR (("clavicle"[MeSH Terms] OR "clavicle"[All Fields] OR "clavicular"[*All Fields*]) AND ("hyperostosis"[MeSH Terms] OR "hyperostosis"[*All Fields*]) AND ("acne vulgaris"[MeSH Terms] OR ("acne"[All Fields] AND "vulgaris"[*All Fields*]) OR "acne vulgaris"[All Fields] OR "acne"[*All Fields*]) AND ("arthritis"[MeSH Terms] OR "arthritis"[*All Fields*]))) OR "Diffuse sclerosing osteomyelitis"[*All Fields*]) OR "Pustulotic arthro-osteitis"[*All Fields*]) OR "Sclerosing osteomyelitis of Garre"[*All Fields*]) OR "Sternocostoclavicular hyperostosis"[*All Fields*]) OR (Sternoclavicular[All Fields] AND pustulotic[All Fields] AND ("osteitis"[MeSH Terms] OR "osteitis"[*All Fields*])) OR "synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome"[*All Fields*]) OR "Garre's osteomyelitis"[*All Fields*]) OR "Diffuse sclerosing osteomyelitis"[*All Fields*]) OR "Pustulotic-arthroosteitis"[*All Fields*]) OR "Lymphoplasmacellular osteomyelitis"[*All Fields*]) OR "Mandibular osteomyelitis"[*All Fields*]) OR (bacterial[All Fields] AND ("osteitis"[MeSH Terms] OR "osteitis"[*All Fields*])) NOT infectious[*All Fields*]) NOT malignant[All Fields] AND "humans"[*MeSH Terms*]) OR "Chronic nonbacterial osteomyelitis"[*All Fields*]) OR (("infertility"[MeSH Terms] OR "infertility"[All Fields] OR "sterile"[*All Fields*]) AND inflammatory[All Fields] AND ("bone diseases"[MeSH Terms] OR ("bone"[All Fields] AND "diseases"[*All Fields*]) OR "bone diseases"[All Fields] OR ("bone"[All Fields] AND "disorder"[*All Fields*]) OR "bone disorder"[*All Fields*]))) OR "autoinflammatory bone disease"[*All Fields*]) OR "recurrent multifocal osteomyelitis"[*All Fields*]) OR "nonbacterial osteomyelitis"[*All Fields*]

- Filters:
  - Language: English
  - Study-type: case series/retrospective/cohort/observational/randomized control trials
  - Age-groups: Adult and Pediatric
  - Full Text Only

- Inclusion Criteria
  - Study Population: Patients (pediatric and adult) with CNO or SAPHO
  - Nature of the Intervention: All study designs (case series/retrospective/cohort/observational/randomized control trials)
  - Case series must have >2 cases
  - Time Period: NO TIME LIMIT
  - Cultural and Linguistic: All languages
  - Full Text available (electronic or hard copy)
  - Includes Outcomes in Core Areas: Death, Life impact, Resource Use/Economical impact, Pathophysiological manifestation

- Exclusion Criteria
  - Patients population is NOT children or adults with CNO or SAPHO
  - Full Text is NOT available
  - Case Reports (< 3 cases)
  - Animal studies
  - Review articles (systematic, literature)

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