

## Defining patient perception of overall well-being and disease activity in the OMERACT Juvenile Idiopathic Arthritis (JIA) core domain set: A report from the JIA working group

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### ABSTRACT

**Objective:** The OMERACT Juvenile Idiopathic Arthritis (JIA) Working Group (WG) aimed to reach agreement on a consensus-based definition and description of the core domain related to patient perception of overall well-being and disease activity.

**Methods:** A committee of patient research partners, clinicians, methodologists, and researchers drafted working definitions and descriptions. The WG conducted two iterative electronic stakeholder surveys to obtain consensus on domain description, definition, and the distinction between patient perception of overall well-being and disease activity. These definitions were then presented at the OMERACT 2023 Special Interest Group (SIG) session for agreement.

**Results:** Forty-five participants, from 7 countries and 4 continents, were comprised of six patients, 18 caregivers, and 21 healthcare providers. The consensus threshold (70%) was exceeded on all survey questions from both stakeholder groups (patients/caregivers, all others). Agreement was obtained on the new definition, description, and domain title, along with agreement on separate assessments of two target domains, patient perception of overall well-being as it relates to disease and patient perception of disease activity.

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**Conclusion:** Through an iterative consensus process and achieving agreement from the OMERACT SIG session attendees, the JIA WG has created a detailed definition and description for the two target domains in the patient perception of overall well-being related to disease core domain of the JIA mandatory core domain set. The next phase of this work will be instrument selection using the OMERACT filter 2.2.

## Introduction

The OMERACT core domain set designed to assess the efficacy of medication in randomized controlled trials (RCTs) for juvenile idiopathic arthritis (JIA), originally established in 1997 [1], was re-evaluated and updated in 2018 by the JIA Working Group (<https://omeract.org/working-groups/juvenile-idiopathic-arthritis/>) to integrate patient and parent/caregiver perspectives into the core domain set. This work culminated in the creation of the OMERACT JIA core domain set recommended for use in RCTs and longitudinal observational studies (LOS). The 2018 established mandatory domains included: pain, joint inflammatory signs, activity limitation/physical function, patient perception of disease (overall well-being), and adverse events including death [2]. After establishing the core domain set, each domain requires a detailed definition and description to serve as the foundation for the instrument selection process and assist in domain matching and content validity as outlined in the OMERACT handbook [3,4].

The JIA Working Group has endeavored to establish potential domain definitions and descriptions for the mandatory core domain of patient perception of disease (overall well-being) through a consensus-based approach with global stakeholders including patients, parents/caregivers, and healthcare providers. This report details the results of this work as presented at the 2023 OMERACT JIA Special Interest Group (SIG) session.

## Methods

The OMERACT JIA Working Group (WG) includes global patients and parent/caregiver representatives, healthcare providers. The JIA Working Group, a voluntary group of individuals, conducted monthly teleconferences from 05/2022–04/2023 to review ongoing work and provide input. The Co-chairs (AC, EMM), patient representatives (BH, GS), OMERACT fellow (EBD), and OMERACT Methodology Advisor (LJM) and Technical Advisory Group member (RC) directed activities.

### Part 1: JIA working group core domain definition and description process

During monthly teleconferences, preliminary domain definitions and descriptions were proposed and revised based on concepts of well-being and health espoused by leading health agencies [5,6] using precedent in pediatric rheumatology [1,7], and expert opinion and consensus within the core working group and guidance from the OMERACT handbook and the Technical Advisory Group. Through input from clinicians and researchers, the group was interested to present the option of two target domains, overall well-being, and disease activity for input and consensus voting.

### Part 2: stakeholder survey 1 & 2

A two round survey process was completed and managed using REDCap electronic data capture tools hosted at Seattle Children's Hospital (SCH) [8,9]. In Italy a paper survey was administered, by choice of the host institution, the remainder were electronic. All data were housed on secure servers at SCH. The first survey was distributed to all known members of the OMERACT JIA Working Group including those who helped develop the preliminary definitions, along with dissemination to patient advocacy groups to increase representation of parent/caregiver/patient voice, including the Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) Parent Work Group (North

America based). Ethics approval obtained through the SCH IRB (STUDY00004250) in the United States, and the Institute for Research and Health Care Istituto Giannina Gaslini ethics board (STUDY 364, 14/04/2020) in Italy. Survey respondents consented to results publication.

The first survey contained four questions in which participants were asked if they agreed with the proposed definition, description, modified title for the domain, and with the creation of the target domains of disease activity and overall well-being. Respondents were asked for edits or suggestions to improve the proposed text. If they agreed to the creation of target domains, a proposed definition of disease activity was presented for review and agreement vote. The steering group members of the Working Group incorporated feedback from the first survey and created a second survey to obtain further consensus on the revised definition and description of the domain based on the first survey. First round survey takers who elected to be re-contacted received the second round survey. Results of the first survey were presented at the beginning of the second survey. Survey instruments are included in supplemental materials (Appendices 1, 2).

### Part 3: OMERACT 2023 special interest group presentation

A summary of the survey results and proposed new definitions was presented at the OMERACT 2023 JIA SIG session on May 2, 2023. All individuals at the OMERACT 2023 conference were invited to join the JIA SIG, and JIA Working Group members not at the conference were invited to join virtually. Present members were asked via an electronic voting platform for their view on agreement on the findings. The work group then presented their planned next steps for instrument selection according to the proposed domain definition and descriptions.

**Table 1**  
Consensus Survey Participant Demographics.

|                  | First Survey (N = 45)<br>n (%) |           | Second Survey (N = 26)<br>n (%) |           |
|------------------|--------------------------------|-----------|---------------------------------|-----------|
| <b>Patient</b>   | 6 (13.3)                       |           | 3 (11.5)                        |           |
| <b>Caregiver</b> | 18 (40.0)                      |           | 10 (38.5)                       |           |
| <b>Other</b>     | 21 (47.0)                      |           | 13 (50.0)                       |           |
|                  |                                | 23        | Health                          | 12        |
|                  |                                |           | Professional                    |           |
|                  |                                | 20        | Researcher                      | 9         |
|                  |                                | 8         | PRO developer                   | 5         |
|                  |                                | 1         | Industry                        | 0         |
|                  |                                | 3         | Advocacy                        | 1         |
|                  |                                | 1         | Other**                         |           |
| <b>Countries</b> | Australia                      | 2 (4.7)   | Australia                       | 1 (4.0)   |
|                  | Canada                         | 3 (7.0)   | Canada                          | 2 (8.0)   |
|                  | Denmark                        | 1 (2.3)   | Denmark                         | 0 (0.0)   |
|                  | Italy*                         | 3 (7.0)   | Italy                           | 2 (8.0)   |
|                  | South Africa                   | 1 (2.3)   | South Africa                    | 1 (4.0)   |
|                  | The Netherlands                | 1 (2.3)   | The Netherlands                 | 1 (4.0)   |
|                  | United States                  | 32 (74.4) | United States                   | 17 (68.0) |

Footnotes: Survey takers were allowed to self-identify and multiple group members identified as multiple categories listed. If a participant identified as both patient representative and healthcare provider, they were counted as a patient representative in the overall percentages presented. \*Indicates paper surveys administered in Italy. \*\*member of the PRCOIN parent working group. 2 individuals in both the primary survey and secondary survey did to identify a country. Pts patients, PRO = patient reported outcomes.

Statistical analysis

Descriptive statistics of the survey results were presented as number and percentage for each major stakeholder group (patient/caregivers and other stakeholders). Consensus threshold was set at a minimum of 70% in each stakeholder group, per OMERACT standards [3]. If a participant identified as both patient representative and healthcare provider, they were counted as a patient representative in the percentages presented.

Results

Results of stakeholder surveys

The first stakeholder survey was completed by 45 individuals: 6 patients, 18 parents/caregivers, and 21 other participants. The first survey was sent to approximately 70 individuals, and we estimate a response rate of 64.3% (45/70). Of note two healthcare providers and four researchers also identified as patient representatives. Seven countries were represented, from four continents (Table 1). Written comments from survey participants were discussed by the Working Group members present at monthly teleconferences; alterations were made to the definition and description of overall well-being and a second survey was created based on group consensus.

The second stakeholder survey was completed by 26 individuals (response rate 66.7% of 39 who allowed recontact), including three patients, 10 parent/caregivers, and 13 other participants including healthcare providers. Of note, healthcare providers could also be identified as patient representatives. Six countries were represented, from four continents. Three responses were left blank on question one, and presumed as no votes. (Table 1)

Survey questions and results included in both stakeholder surveys are available in Table 2 and the supplemental material. Across all questions, a consensus greater than 70% was reached both by patient

Table 2  
Iterative stakeholder survey results:

| Participants were asked if they agreed with the following proposals |  | Patient/<br>Caregiver<br>(N = 24) | Others<br>(N = 21) |
|---|--|-----------------------------------|--------------------|
| <b>Survey 1</b>   |  | <b>Percent Agreement</b>          |                    |
| Q1<br>Definition  | <b>Patient perception of disease (overall well-being)</b> considers the personal view of the patient on all the ways that JIA affects the patient, including their sense of overall health, their health-related quality of life, and their perception of how well their disease activity is controlled.   | 100%                              | 95.2%              |
| Q2<br>Description   | <b>Patient perception of disease (overall well-being)</b> captures the personal perception of the patient of the aspects of the patient's life impacted by the patient's JIA. It includes the level of disease activity, burden of illness on health (physical, emotional, mental, or social), and impact of treatment/medications (positive effects or negative health symptoms, administration of medication).   | 95.8%                             | 85.7%              |
| Q3<br>Do you agree with Option 1?                                   | <b>Option 1: Patient perception of disease (overall well-being)</b> domain consists of an overarching measure of overall well-being. <b>Patient (or parent/caregiver) perception of disease activity should also be measured independently.</b>  | 79.2%                             | 71.4%              |
| Q3<br>Description   | <b>Option 2: Patient perception of disease (overall well-being)</b> domain consists of an overarching measure of overall well-being. <b>Patient perception of disease activity is part of this domain and should not be assessed separately.</b>   | 89.5%                             | 80%                |
| Q4<br>Title   | <b>Patient perception of disease activity</b> includes the aspects of the disease that in the personal judgment of the patient (or parent/caregiver) <b>directly</b> relate to the JIA (such as joint swelling, pain, stiffness), and may possibly respond to treatment/medication.<br><br>This assessment would not include the following: Symptoms or physical findings thought to be due to another disease process or diagnosis, or due to treatment, or due to irreversible damage from JIA.<br><br>To clarify the title (name) of the domain to better match the definition, we propose a change from <b>"Patient perception of disease (overall well-being)"</b> to <b>"Patient perception of overall well-being as relates to disease (JIA)"</b> | 100%                              | 100%               |
|   |  | <b>N = 13</b>                     | <b>N = 13</b>      |
| <b>Survey 2</b>   |  |                                   |                    |
| Q1<br>Definition  | <b>Patient perception of overall well-being as relates to disease (JIA)</b> considers the personal view of the patient (or parent/caregiver) on all the ways that JIA affects the patient, including their sense of overall health.  | 84.6%*                            | 92.3%<br>**        |
| Q2<br>Description   | <b>Patient perception of overall well-being as relates to disease (JIA)</b> captures the personal view (of the patient or parent/caregiver) of the aspects of the patient's life impacted by the patient's juvenile idiopathic arthritis. It may be affected by the level of disease activity, burden of illness on health (physical, emotional, mental, or social), and impact of treatment/medications (positive effects or negative health symptoms (e.g. side effects), administration of medication).   | 100%                              | 100%               |

Footnotes: If participants identified as both patient representative and healthcare provider, they were counted as a patient representative in the overall percentages presented. \*2 responses were left blank and assumed as no votes, \*\*1 response was left blank and assumed as a no vote.

representatives and other participants.

1.%2 Results from OMERACT 2023 JIA SIG session

At the OMERACT 2023 JIA SIG session the consensus work was presented. Nineteen participants were present, including three patient representatives, and 16 healthcare providers, 18 of whom voted. 94.4% (17/18) agreed with our findings. A second question was posed, "Do you agree that we need separate measurement tools for overall well-being and for disease activity?".89% (N = 16/18) agreed with creation of target domains for instrument selection.

Consensus-based domain title, definitions, and descriptions

Through this consensus work including discussion within the JIA Working Group, two iterative stakeholder surveys, and discussion at the OMERACT 2023 SIG session a new domain title, definition, and descriptions for overall well-being and disease activity were created. The consensus new domain title is "Patient perception of overall well-being related to disease (JIA)". The new domain definition is "Patient perception of overall well-being as relates to disease (JIA) considers the view of the patient (or parent/caregiver) on all the ways JIA affects the patient, including their sense of overall health". The new consensus definitions and descriptions are available in tables 3 and 4. Further, the separation into target domains of overall well-being and disease activity for the purpose of measurement and instrument selection was agreed upon. These changes resulted in modification of the "OMERACT onion" of the JIA Core Domain Set domains. Fig. 1

Discussion

Through iterative consensus-based work within the JIA Working Group, two stakeholder surveys, and discussion and agreement at the

**Table 3**  
OMERACT Domain definition and description report template. Agreed upon domain definition, description and title – Patient Perception of Overall Well-being.

|  |  |  |  |
|--|--|--|--|
| <b>Working Group: Juvenile Idiopathic Arthritis (JIA)</b>  |  | <b>Date completed: 2023-05-23</b>        |  |
| <b>Population:</b><br>Patients with JIA  | <b>Intervention(s):</b> Pharmacological and non-pharmacological interventions for JIA  | <b>Control(s):</b><br>placebo/drug       | <b>Context:</b> Randomized control trials and longitudinal observational studies   |
| <b>What is the name that you give to your target domain?</b>   | <b>Patient perception of overall well-being</b>  | <b>Is this part of a broader domain?</b> | <input type="checkbox"/> No<br><input checked="" type="checkbox"/> Yes ....If yes, which one ___ Patient perception of overall well-being related to disease (JIA) |
| <b>Tell us more about that domain</b> –this is your domain definition. what is the breadth, depth – what do you want to be able to see.  | <b>Domain Definition:</b> Patient perception of overall well-being as relates to disease (JIA) considers the view of the patient (or parent/caregiver) on all the ways JIA affects the patient, including their sense of overall health.   |  |  |
|  | <b>Domain description overall well-being:</b> Patient perception of overall well-being as relates to disease (JIA) captures the view (of the patient or parent/caregiver) of the aspects of the patient’s life impacted by the patient’s JIA. It may be affected by the level of disease activity, burden of illness on health (physical, mental, or social), and impact of treatment/medications (positive changes, (e.g. improved mobility), or negative changes (e.g. side effects and administration of medication).<br><br>Overall wellbeing is an important area and our group found that two domains needed to be measured in a core outcome set. Overall wellbeing as it relates to the way the disease is impacting the life of a person with JIA (this current definitional template) and equally important Overall wellbeing as it relates to disease activity.<br>-Please look to the Patient perception of overall wellbeing as it relates to disease activity for the complementary definition. (Table 4)  |  |  |
| Which of the <b>core areas</b> does this fall into? (check one)  | <input type="checkbox"/> Pathophysiological manifestations<br><input type="checkbox"/> Death/Lifespan<br><input checked="" type="checkbox"/> Life impact<br><input type="checkbox"/> Resource use (i.e., costs)  |  |  |
| <b>Tracking for future reference.... (we suggest you track these now while you are thinking about it, but they are not mandatory. You (or your successors) will need them later in instrument selection)</b> |  |  |  |
| <b>...How did you come to understand this target domain well?</b>  | <p><i>Qualitative findings (i.e., add relevant quotes from patients, stakeholders that aid in understanding of what this is....and what it is not)</i></p> <p>“ I would consider changing the final phrase of the definition from "affects the patient's sense of overall health" to "affects the patient's overall health." I like that the definition is shortened, but doing so places more emphasis on the "perception," "personal view," and "sense of" in the definition while removing context that shows the importance of the patient perspective. It could be perceived as downplaying/negating the patient perspective. Far too often, patients feel like their perspective is dismissed, particularly when clinically assessed disease activity is low, and having 3 qualifiers feels dismissive rather than empowering/collaborative. Alternately, taking out the "personal" from "personal view of the patient" in the definition while leaving "personal view" in the description might have the same effect.”</p> <p>“Use consistent language to describe positive and negative impact of treatment/medications”</p> <p>“Trying to smooth it out and thinking about health care literacy. This may include amount of disease activity, difficulties due to the disease (physical, emotional or social) and its treatment (good and bad things about the treatment such as side effects).”</p> <p>“I would have omitted "how well their disease is controlled" as this seems to be a different construct (disease activity)”</p> <p><i>Other available definitions, frameworks used (i.e., did you take definition from another framework, or another working group – both are great if they work for you – cite here)</i></p> <p>We reviewed descriptions of concepts of overall well-being and health in publications of public health organizations, and reviewed items measuring overall wellbeing in widespread use to launch workgroup discussions <sup>5, 6, 7</sup></p> |  |  |
| <b>...Are there any “it all depends” type factors. Factors that make a difference in the number/score obtained?</b>  | <p><i>Examples: +/- use of assistive device, type of imaging machine, technician variability, time of day...</i></p> <ul style="list-style-type: none"> <li>• Scores reported directly from patients versus proxy (e.g., parent) may vary</li> <li>• Existence of co-morbid conditions could make it difficult for respondent to disentangle impacts on overall well-being of JIA vs other conditions</li> <li>• Length of reporting period</li> </ul>   |  |  |

2023 OMERACT conference the JIA Working Group has produced a new title, detailed descriptions, and definitions for the core domain of patient perception of overall well-being related to disease (JIA). Patient and parent/caregivers and the healthcare providers alike agreed on these domain definitions and descriptions along with the creation of two target domains including overall well-being and disease activity for the

purpose of instrument selection.

This work may be limited by the number of participants in the working group meetings and at the JIA SIG, which may limit our representation. The next steps for this work will be the instrument selection phase using the evidence-based framework of the ‘OMERACT Filter Instrument Selection Algorithm’ most recently updated in 2021 [10], and

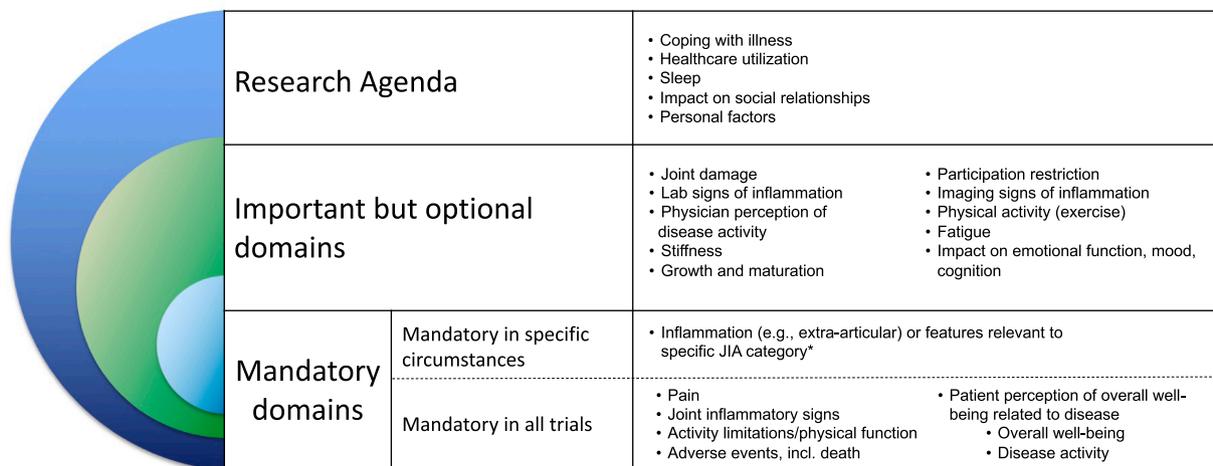
**Table 4**

OMERACT Domain definition and description report template. Agreed upon domain definition, description and title – Patient Perception of Disease Activity (See reference [12]).

|  |  |  |  |
|--|--|--|--|
| <b>Working Group: Juvenile Idiopathic Arthritis (JIA)</b>  |  | <b>Date completed: 2023-05-23</b>        |  |
| <b>Population:</b><br>Patients with JIA  | <b>Intervention(s):</b> Pharmacological and non-pharmacological interventions for JIA  | <b>Control(s):</b><br>placebo/drug       | <b>Context:</b> Randomized control trials and longitudinal observational studies   |
| <b>What is the name that you give to your target domain?</b>   | <b>Patient perception of disease activity</b>  | <b>Is this part of a broader domain?</b> | <input type="checkbox"/> No<br><input checked="" type="checkbox"/> Yes ....If yes, which one Patient perception of overall well-being related to disease (JIA) |
| <b>Tell us more about that domain</b> –this is your domain definition. what is the breadth, depth – what do you want to be able to see.  | <b>Domain Definition:</b> Patient perception of overall well-being as relates to disease (JIA) considers the view of the patient (or parent/caregiver) on all the ways JIA affects the patient, including their sense of overall health.   |  |  |
|  | <b>Domain description disease activity:</b> Patient perception of disease activity includes the aspects of the disease that in the personal judgment of the patient (or parent/caregiver) directly relate to the JIA (such as joint swelling, pain, stiffness), and may possibly respond to treatment/medication.<br>This assessment would not include the following: symptoms or physical findings thought to be due to another disease process or diagnosis, or due to treatment, or due to irreversible damage from JIA.<br><br>Overall well-being is an important area and our group found that two domains needed to be measured in a core outcome set. Overall well-being as it relates to disease activity (this current definitional template) and equally important overall well-being as it related to the way the disease is impacting the life of a person with JIA -Please look to the Patient perception of overall well-being as it related to the way the disease is impacting the life of a person with JIA for the complementary definition. (Table 3)   |  |  |
| Which of the <b>core areas</b> does this fall into? (check one)  | <input checked="" type="checkbox"/> Pathophysiological manifestations<br><input type="checkbox"/> Death/Lifespan<br><input type="checkbox"/> Life impact<br><input type="checkbox"/> Resource use (i.e., costs)  |  |  |
| <b>Tracking for future reference.... (we suggest you track these now while you are thinking about it, but they are not mandatory. You (or your successors) will need them later in instrument selection)</b> |  |  |  |
| <b>...How did you come to understand this target domain well?</b>  | <p><i>Qualitative findings (i.e., add relevant quotes from patients, stakeholders that aid in understanding of what this is...and what it is not)</i></p> <p>“I think that many things could "directly relate to the JIA" without having anything to do with the amount of inflammation in the patient's joints, eyes, skin, etc. I would state that those aspects of the disease that ... directly relate to the amount of inflammation the patient is currently experiencing.”</p> <p>“Consider including the idea that these things might be reversible - "...may possibly respond to treatment/medication and might be reversed.”</p> <p>“he examples of Pain, Swelling and stiffness are core but I wonder if there should be mention of subtype specific morbidity such as uveitis, rash, fever and (slightly more difficult is the concept of fatigue, which can certainly be a manifestation of disease activity but is also often not necessarily attributable to disease activity)”</p> <p>“If a patient has other symptoms or perceives to have other JIA related symptoms, I believe it would be helpful to discuss. JIA has so many subsets and to not include other diagnosis may cause other issues to be overlooked.”</p> <p><i>Other available definitions, frameworks used (i.e., did you take definition from another framework, or another working group – both are great if they work for you – cite here)</i></p> <ul style="list-style-type: none"> <li>No other frameworks or definitions were used in the creation of this definition and description</li> <li>There is a consensus effort amongst researchers to define physician global assessment of disease activity which informed the discussion.<sup>12</sup></li> </ul> |  |  |
| <b>....Are there any “it all depends” type factors. Factors that make a difference in the number/score obtained?</b>   | <p><i>Examples: +/- use of assistive device, type of imaging machine, technician variability, time of day...</i></p> <ul style="list-style-type: none"> <li>Scores reported directly from patients versus proxy (e.g., parent) may vary</li> <li>Scoring may consider extra-articular manifestations in a single item vs multiple items</li> </ul>   |  |  |

the OMERACT handbook for guidance [3]. One example candidate instrument is the JAMAR composite tool which contains individual disease activity and overall well-being items. [11] Candidate instruments such as this will be put through the filter, using the c detailed description of the domain as the primary step for candidate instrument domain match.

Use of this process will allow for robust and accurate instrument selection leading to reliable measurement of the two target domains of overall well-being and disease activity



**Fig. 1.** OMERACT domain framework for juvenile idiopathic arthritis (JIA) studies including mandatory domains in specific circumstances. Recommended for use. \*Illustrative examples (incomplete list): oligoarthritis: eye inflammation/uveitis; systemic JIA: fever, systemic rash, macrophage activation syndrome features; psoriatic arthritis: psoriasis; enthesitis-related arthritis: enthesitis, etc.

**Conclusion**

Through consensus, the OMERACT JIA Working Group has created a clear and detailed definitions and descriptions for the two target domains for the core domain of Patient Perception of Overall Well-being Related to Juvenile Idiopathic Arthritis and will next move on to the instrument selection phase of this work.

**Declaration of Competing Interest**

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests:

Erin Balay-Dustrude reports travel was provided by Omeract - Outcome Measures in Rheumatology. Ben Horgan reports travel was provided by Omeract - Outcome Measures in Rheumatology. Grayson Shultz reports travel was provided by Omeract - Outcome Measures in Rheumatology. Alessandro Consolaro reports a relationship with Pfizer that includes: funding grants and speaking and lecture fees. Joost F. Swart reports a relationship with Amgen Inc that includes: consulting or advisory. Esi M Morgan reports a relationship with Pfizer that includes: funding grants. Esi M Morgan reports a relationship with Agency for Healthcare Research and Quality that includes: funding grants. Esi M Morgan reports a relationship with American College of Rheumatology that includes: speaking and lecture fees and travel reimbursement. Esi M Morgan reports a relationship with Pediatric Rheumatology Care and Outcomes Improvement Network that includes: board membership.

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**Author contribution statements**

All listed authors have made substantial contributions to this work including meeting ICJME requirements for authorship

**Supplementary materials**

Supplementary material associated with this article can be found, in the online version, at [doi:10.1016/j.semarthrit.2023.152340](https://doi.org/10.1016/j.semarthrit.2023.152340).

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