

Prioritized Agenda for Mental Health Research in Pediatric Rheumatology from the Childhood Arthritis and Rheumatology Research Alliance Mental Health Workgroup

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ABSTRACT. **Objective.** Mental health problems are prevalent in youth with rheumatologic disease. Gaps in knowledge exist regarding their effect, as well as strategies for detection and effective treatment. To address these gaps, the Childhood Arthritis and Rheumatology Research Alliance (CARRA) Mental Health Workgroup developed and prioritized an agenda of research topics.

Methods. We systematically reviewed the literature and identified 5 major research domains in further need of study: (A) mental health burden and relationship to pediatric rheumatologic disease, (B) effect of mental health disorders on outcomes, (C) mental health awareness and education, (D) mental health screening, and (E) mental health treatment. Research topics within these areas were developed by workgroup leaders and refined by the workgroup. Members were surveyed to prioritize the topics by importance, feasibility of study, and actionability.

Results. Fifty-nine members (57%) completed the survey. Among the proposed research topics, 31/33 were rated as highly important and 4/33 were rated highly for importance, feasibility, and actionability. Topics rated most important related to (A) mental health burden and relationship to rheumatologic disease, and (B) the effect of mental health on outcomes. Topics rated most feasible and actionable were related to (D) mental health screening.

Conclusion. Addressing gaps in knowledge regarding mental health in youth with rheumatologic disease is essential for improving care. We have identified high priority research topics regarding mental health of pediatric rheumatology patients in need of further investigation that are feasible to study and believed to lead to actionable results in patient care.

Key Indexing Terms: juvenile dermatomyositis, juvenile idiopathic arthritis, mental health, pediatrics, systemic lupus erythematosus

Depression, anxiety, and other mental health symptoms are common in children and adolescents with rheumatologic diseases, affecting at least 15–40%¹. However, significant deficiencies exist in access to mental health providers and the identification and treatment of these symptoms within pediatric

rheumatology clinics^{2,3}. Similar to youth with other chronic disease, evidence suggests that mental health symptoms in youth with rheumatologic disease are associated with poorer quality of life^{4,5}, poor medication adherence⁶, and possibly worse disease outcomes⁷. Parents and siblings of youth with rheumatologic

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diseases also experience mental health symptoms, which are likely associated with their own adjustment to the affected child's illness and associated mental and physical symptoms⁸. Despite a growing appreciation that these symptoms are prevalent and detrimental, our understanding of the complex interplay between mental health symptoms and rheumatologic diseases is nascent. It is often unclear whether these symptoms arise as part of the inflammatory disease process itself, whether they are a reaction to the disease, or whether they are coincident. Further, studies to assess strategies for adequate identification and effective treatment of mental health symptoms in this population are lacking¹.

Our prior review of the literature on mental health symptoms in youth with rheumatologic diseases found that small, single-center, cross-sectional studies represent the bulk of evidence in the field and indicate that mental health symptoms are prevalent in this population^{1,9}. These studies primarily focus on juvenile idiopathic arthritis (JIA) and systemic lupus erythematosus (SLE) and are limited in their representation of minority patients and sociodemographic information. Few longitudinal studies have been conducted. Thus, significant gaps in our knowledge exist regarding risk factors for mental health symptoms, their longterm effect, strategies for mental health education, best screening practices, and effective treatment^{1,9}. Few studies addressed potential roles for improving mental health care through integration of mental health services and educational opportunities for rheumatologists, despite growing evidence pointing to efficacy in general pediatrics and other pediatric chronic disease, such as diabetes and cystic fibrosis^{10,11}. While our comprehension of the interactions between mental health and physical health in youth with chronic disease is expanding, the complicated relationship between mental health, inflammatory disease, and its treatment is less well understood. Gaps in our knowledge exist regarding the potentially unique mental health needs of youth with rheumatologic disease, and optimal screening and treatment strategies for this population. Thus, to help advance our knowledge and coordinate the efforts of investigators committed to mental health research in pediatric rheumatology, the Mental Health Workgroup for the Childhood Arthritis and Rheumatology Research Alliance (CARRA) aimed to develop a prioritized research agenda. CARRA is an international research collaborative of pediatric rheumatologists, allied health professionals, patients, parents, and research associates. CARRA includes > 550 pediatric rheumatology researchers and > 95% of pediatric rheumatologists in the United States and Canada.

Prioritized research agendas promote clinically important and actionable research across disciplines to address areas of need^{12,13,14}. These are developed through literature reviews, in-person discussions, membership surveys, and Delphi methods to obtain consensus^{13,14,15}. The intent of developing a prioritized research agenda was to promote clinically relevant, important, and feasible research efforts that address gaps in knowledge related to the mental health of youth with rheumatologic disease,

and that are likely to lead to changes in practice and improvements in care.

MATERIALS AND METHODS

Prioritization methodology was adapted from the Prioritization Criteria Methods (PiCMe) of the US Agency for Healthcare Research and Quality (AHRQ). This methodology includes (1) systematically reviewing the literature, (2) determining gaps in evidence, (3) developing research questions from evidence gaps, (4) ranking research questions, and (5) publishing research needs¹⁶.

Systematic review of the literature on mental health in pediatric rheumatology was completed and published by members of the CARRA Mental Health Workgroup^{1,9}. Through iterative discussions occurring over conference calls, a taskforce of 4 workgroup members (AD, AK, MR, TR) reviewed the findings and developed consensus on 5 major research domains in need of further study: (A) mental health burden and relationship to pediatric rheumatologic disease, (B) effect of mental health disorders on outcomes, (C) mental health awareness and education, (D) mental health screening, and (E) mental health treatment. The taskforce then developed a list of 36 draft research topics within these domains based on the main identified knowledge gaps. These topics were discussed, refined, and revised at the CARRA Mental Health Workgroup Annual Meeting in April 2018, which 59 workgroup members attended, including pediatric rheumatologists, psychologists, social workers, patients, parents, research coordinators, and industry members. Additional discussion and collection of feedback were conducted through e-mail and a subsequent conference call to enable participation from workgroup members missing in attendance from the CARRA Annual Meeting. The resulting product of the iterative discussion and revision process was a list of 33 refined research topics that fell within the 5 research domains (Table 1). An online survey was created to prioritize the 33 topics based on 3 fields adapted from the AHRQ PiCMe: importance, feasibility, and actionability. Importance was defined as the relevance of the topic to advancing clinical care and research, considering existing knowledge. Feasibility was defined as the ease with which research could be conducted and completed. Actionability was defined as the ability to apply the results of research toward advancing clinical care.

The survey was distributed to all members of the workgroup e-mail listserv (n = 103). Respondents could choose to opt out of the survey (i.e., if not willing to participate or owing to conflict of interest). Demographic and affiliation information was collected. We categorized participants into 3 subgroups of interest: more experienced (≥ 10 yrs in rheumatology) versus less experienced (< 10 yrs) members; rheumatologists (rheumatology attendings and rheumatology fellows) versus non-rheumatologists (all other workgroup members); and among rheumatologists, research-focused ($\geq 50\%$ full-time effort for research) versus not.

Respondents were asked to score each research topic on a 5-point scale for the 3 fields: importance, feasibility, and actionability (1 = low, 2 = somewhat low, 3 = neutral, 4 = somewhat high, 5 = high). Mean ranking scores were calculated. Research topics achieving mean ranking scores of ≥ 4 for importance, feasibility, or actionability were deemed "highly" important, feasible, or actionable, respectively. Additionally, members were asked to provide an overall ranking (in consideration of importance, feasibility, and actionability) of research topics within each research domain.

This study was approved by The Children's Hospital of Philadelphia Institutional Review Board (IRB 18-015073) and survey responses were obtained with the participants' informed consent.

Statistical analysis. Descriptive statistics were used to determine the mean and SD of response scores for each research topic. Mean ranking scores and SD were also calculated for the overall ranking of each of the different topics within domains to determine the highest-ranked research topics for

Table 1. Topics for mental health research in pediatric rheumatology by research domain defined by the CARRA Mental Health Workgroup.

Domain	Research Topic
(A) Mental health burden and relationship to pediatric rheumatologic disease	<p>(A1) Determine the prevalence and incidence of mental health disorders in pediatric patients with rheumatologic disease, as well as sociodemographic and disease-specific risk factors.</p> <p>(A2) Determine the relationship between mental health disorders, disease onset, and disease course.</p> <p>(A3) Investigate the relationship between mental health and rheumatologic disease treatments.</p> <p>(A4) Investigate the biologic basis of mental health disorders.</p> <p>(A5) Examine the effect of rheumatologic disease on neuropsychological development.</p>
(B) Effect of mental health disorders on outcomes	<p>(B1) Investigate the effect of mental health on clinical outcomes, such as disease activity.</p> <p>(B2) Investigate the effect of mental health on longterm clinical outcomes, such as disease damage and mortality.</p> <p>(B3) Investigate the effect of mental health on health-related behaviors, such as medication adherence.</p> <p>(B4) Determine the effect of mental health on outcomes related to transition to adult care.</p> <p>(B5) Investigate the effect of mental health on healthcare use and costs.</p> <p>(B6) Investigate the effect of mental health on quality of life, social outcomes, education attainment, and work functioning.</p>
(C) Mental health awareness and education	<p>(C1) Understand the level of awareness for importance of mental health among groups.</p> <p>(C2) Determine the impact of mental health education of patients/families on their perceived acceptability of mental health intervention.</p> <p>(C3) Determine the impact of mental health education of rheumatology clinicians on their perceived feasibility/acceptability, and implementation of mental health practices/intervention.</p> <p>(C4) Define gaps in knowledge for community behavioral health providers pertaining to specific mental health needs of patients with childhood-onset rheumatologic disease and their families.</p> <p>(C5) Determine the impact of education on policy change for compensation of mental health screening/intervention.</p>
(D) Mental health screening	<p>(D1) Determine which mental health conditions are most important to screen.</p> <p>(D2) Determine the accuracy of mental health screening tools for identifying mental health conditions in specific pediatric rheumatology disease populations.</p> <p>(D3) Determine the optimal timing, settings, and process for mental health screening.</p> <p>(D4) Determine barriers and facilitators to mental health screening in the pediatric rheumatology setting.</p> <p>(D5) Determine acceptability of mental health screening in pediatric rheumatology clinics for patients, caregivers, clinicians, and identify strategies to improve acceptability.</p> <p>(D6) Determine the feasibility and sustainability of mental health screening in the pediatric rheumatology setting, including cost-effectiveness, ethical, and legal aspects.</p> <p>(D7) Determine the impact of mental health screening on acceptability of mental health treatment for patients/caregivers.</p> <p>(D8) Determine the efficacy of mental health screening in the pediatric rheumatology setting.</p> <p>(D9) Determine the relationship between mental health screening and disease-related outcomes.</p>
(E) Mental health treatment	<p>(E1) Define barriers and facilitators to mental health treatment.</p> <p>(E2) Determine whether adjustment/coping interventions around the time of diagnosis prevent the onset of major depression/anxiety or affect disease-related outcomes.</p> <p>(E3) For different populations and different mental health disorders, determine which mental health treatment modalities are most efficacious.</p> <p>(E4) Investigate the role of immunosuppressive therapy in treating mental health conditions in pediatric patients with rheumatologic disease.</p> <p>(E5) Determine effective mental health treatment delivery options that differ by mode, provider, and setting.</p> <p>(E6) Investigate factors contributing to sociocultural disparities in mental health care and test interventions to reduce disparities.</p> <p>(E7) Determine whether parent/caregiver-specific interventions improve mental health and disease-related outcomes.</p> <p>(E8) Investigate the cost-effectiveness of mental health treatment in pediatric rheumatology setting.</p>

Listed here are the 33 refined research topics defined by the Childhood Arthritis and Rheumatology Research Alliance (CARRA) Mental Health Workgroup members. Bolded research topics are the topics that achieved the highest means for overall ranking of research topics within each domain. All research topics achieved mean “importance” ranking scores of ≥ 4 on a 5-point scale (1 = low, 2 = somewhat low, 3 = neutral, 4 = somewhat high, 5 = high), except for (E1) with mean score 3.9 and (E4) with mean score 3.8.

each of the 5 domains. Statistical analysis was performed using Stata 14 (StataCorp.).

We conducted 3 comparison analyses, by experience level, rheumatologist status, and level of research focus. To compare scores by each of these characterizations, we constructed mixed effects linear regression models with each field (importance, feasibility, actionability) as an outcome and with each subgroup characterization as a predictor. Random effects were specified for the respondent-specific intercept to account for intrarespondent correlations in the scores. Because patients/parents and mental health providers were 2 specific groups that were underrepresented in the workgroup membership and survey, we highlighted their responses on topic and domain importance and overall rankings of topics.

RESULTS

Participant characteristics. Seventy-three participants (71%) responded to the survey: 59 participants (57%) completed the survey, 6 participants (6%) opted out, and 8 participants (8%) started but did not complete the survey. The majority of respondents were pediatric rheumatologists from academic centers, corresponding to the overall composition of the workgroup and CARRA membership. A relatively even distribution of members represented small centers (1–3 rheumatologists), medium centers (4–8), and large centers (9–14), and 53% of members were early-career (< 10 yrs of experience). Among rheumatologists, 38% were research-focused. Non-rheumatologist participants included other medical providers, mental health providers, patients and parents, research coordinators, a sociologist, a member of industry, and a health educator (Table 2).

Highly important research topics. Among the proposed research topics, 31/33 were rated as highly important (≥ 4 mean score) by the workgroup and all were rated relatively highly with mean scores ranging from 3.8–4.7 out of 5. Results of mixed effects models showed that none of the subgroups were independent predictors of importance scores. Overall, importance scores were similarly high in more and less experienced members [$\beta(\text{experienced}) = 0.11, p = 0.4$], rheumatologists and non-rheumatologists [$\beta(\text{experienced}) = 0.05, p = 0.7$], and research-focused rheumatologists and other rheumatologists [$\beta(\text{experienced}) = 0.12, p = 0.8$]. The highest scored topics for importance were in Domain A (mental health burden and relationship to pediatric rheumatologic disease, mean \pm SD = 4.5 \pm 0.7) and Domain B (effect of mental health disorders on outcomes, mean \pm SD = 4.5 \pm 0.7). These 2 domains achieved the highest mean importance scores in each member subgroup. Of specific interest in the non-rheumatologist subgroup were the 4 patient/parent representatives and the 3 mental health providers. Among patients/parents, the highest mean importance scores across topics were in Domain A (mean \pm SD = 4.8 \pm 0.3) and Domain D (mental health screening, mean \pm SD = 4.7 \pm 0.4). Among mental health providers, the highest mean importance scores were in Domain B (mean \pm SD = 4.9 \pm 0.2) and Domain D (mean \pm SD = 4.7 \pm 0.2).

Highly feasible research topics. Regarding feasibility, 4/33 achieved

Table 2. Demographics and characteristics of the CARRA Mental Health Workgroup members participating in the research agenda prioritization survey.

	CARRA Mental Health Workgroup Members Participating in Prioritization Survey, n = 59
Sex	Female: 49 (83) Male: 10 (17)
Age	20–29: 1 (2) 30–39: 20 (51) 40–49: 18 (31) 50–59: 7 (12) 60–69: 3 (5)
Affiliation	Rheumatologist: 42 (71) Nurse practitioner: 2 (3) Research coordinator: 6 (10) Parent/patient: 4 (7) Behavioral health provider*: 3 (5) Other**: 4 (6)
Rheumatology experience, yrs, n = 56	0–4: 24 (43) 5–9: 11 (20) 10–19: 15 (27) 20–35: 6 (11)
Rheumatology setting, n = 42	Academic: 37 (63) Hospital-based: 3 (7) Private practice: 1 (2) Government: 1 (2)
Rheumatologist focus, n = 42	Clinical: 26 (62) Research: 16 (38)
Rheumatologists per practice, n = 56	1–3: 16 (29) 4–8: 25 (35) 9–14: 15 (27)

Values are n (%). * Behavioral health provider: psychologist (2), social worker (1). ** Other: nephrologist (1), industry (1), sociologist (1), and nonprofit health educator. CARRA: Childhood Arthritis and Rheumatology Research Alliance.

mean scores ≥ 4 and determined to be highly feasible (mean scores ranged from 2.9–4.2). There were no significant differences seen between subgroups, though experienced members tended to give lower scores than less experienced ones [$\beta(\text{experienced}) = -0.22, p = 0.1$] and research-focused rheumatologists tended to give higher scores than other rheumatologists [$\beta(\text{research-focused}) = 0.23, p = 0.2$]. Overall, Domain D (mental health screening) topics were rated as most feasible (mean \pm SD = 3.9 \pm 0.9). This domain achieved the highest mean feasibility scores in each member subgroup.

Highly actionable research topics. Regarding actionability, 6/33 were rated as highly actionable (mean scores ranged from 3.4–4.2), including all 4 of the topics rated as highly feasible (Figure 1). Overall, experienced members gave significantly lower actionability scores than less experienced members [$\beta(\text{experienced}) = -0.33, p = 0.02$], while actionability scores between rheumatologists and non-rheumatologists were similar

$[\beta(\text{rheumatologist}) = -0.002, p = 1.0]$. Research-focused rheumatologists tended to give higher actionability scores compared to other rheumatologists, but this difference was not statistically significant $[\beta(\text{research-focused}) = 0.23, p = 0.2]$.

Overall, the topics rated as most actionable were in Domain D (mental health screening, mean \pm SD = 4.0 ± 0.9). Mean actionability scores were highest for Domain D in each subgroup except non-rheumatologists, who rated Domains A through D equally high (all with mean scores \pm SD = 3.9 ± 1.0).

Summary of highest priority research topics. The 4 research topics achieving mean scores of ≥ 4 in all fields and thus determined to be highly important, feasible, and actionable were (in order of highest to lowest mean importance score): (A1) to determine the prevalence and incidence of mental health disorders in pediatric patients with rheumatologic disease, as well as sociodemographic and disease-specific risk factors; (D1) to determine which mental health conditions are most important to screen; (D2) to determine the accuracy of mental health screening tools specifically for pediatric rheumatology disease populations; and (D4) to determine the barriers and facilitators to mental health screening in the pediatric rheumatology setting (Figure 1).

For overall ranking priorities of research topics within each research domain, the top 2 for each of the 5 domains are shown bolded in Table 1. The priority rankings within domains by the overall workgroup yielded the same top 2 topics when we examined each subgroup, except for the subgroup of research-focused

rheumatologists in the domains of (B) effect of mental health and (E) mental health treatment. Among research-focused rheumatologists, the topic (B2) “investigate the impact of mental health on longterm clinical outcomes” ranked higher than (B3) “investigate the impact of mental health on health-related behaviors;” and the topic (E6) “investigate factors contributing to sociocultural disparities in mental health care” outranked (E2) “determine whether adjustment/coping interventions around the time of diagnosis prevent the onset of major depression/anxiety or affect disease-related outcomes.”

Rankings by mental health providers yielded similar results to the overall group, with the exception of the domain of mental health education, in which they ranked topic (C3) “determine the impact of mental health education of rheumatology clinicians” above topic (C1) “understand the level of awareness for the importance of mental health among stakeholder groups.” Patients/parents ranked both of these topics as highest priority in this domain. Further differences among patients/parents to the overall group were in the domain of (A) mental health burden, in which they prioritized topic (A3) “investigate the relationship between mental health and rheumatologic disease treatments;” in the domain of (B) mental health effect, where they prioritized topic (B2) “investigate the impact of mental health on longterm clinical outcomes;” and in the domain of (D) mental health screening, in which they prioritized topic (D4) “determine barriers and facilitators to mental health screening in the pediatric rheumatology setting.”

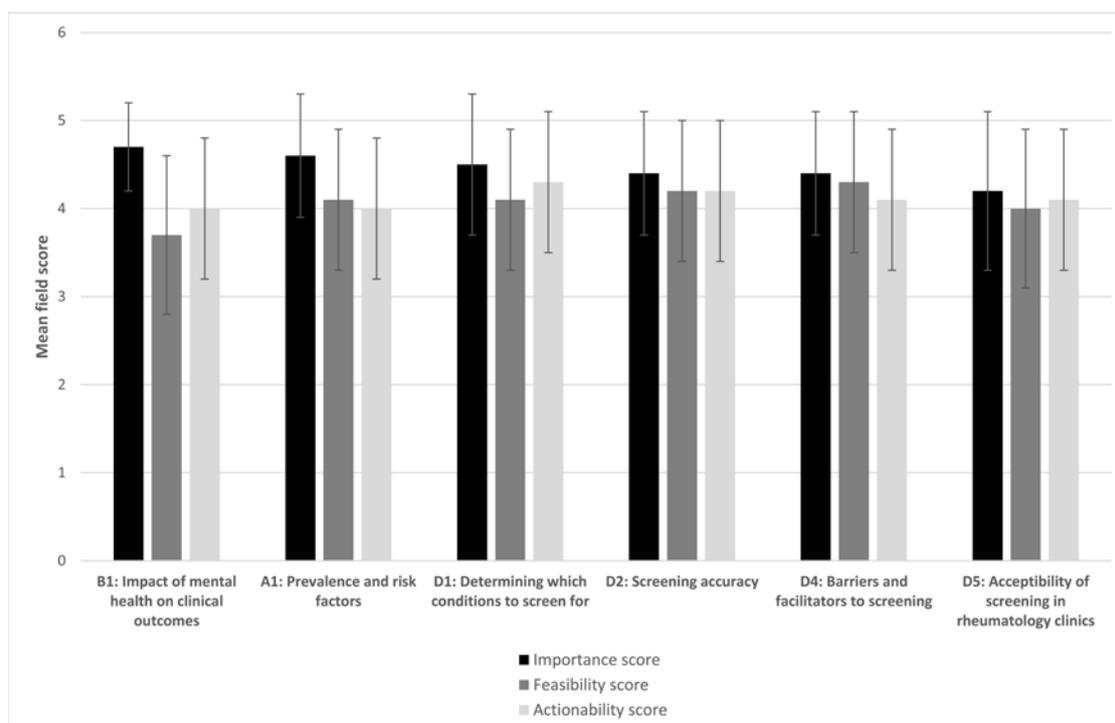


Figure 1. Research topics shown achieved ≥ 4 mean scores for actionability. Importance, feasibility, and actionability were scored on a 5-point scale (1 = low, 2 = somewhat low, 3 = neutral, 4 = somewhat high, 5 = high). Mean scores and SD are shown.

DISCUSSION

We prioritized a research agenda for the CARRA Mental Health Workgroup by surveying a majority of workgroup participants to coordinate and promote research efforts in the most significant areas defined by researchers, providers, and families with a vested interest in improving efforts in mental health care for youth with rheumatologic diseases. Four research topics were determined to be highly important, highly actionable, and highly feasible. These topics were (1) determine the prevalence and incidence of mental health disorders in pediatric patients with rheumatologic disease; (2) determine which mental health conditions are most important to screen; (3) determine the accuracy of mental health screening tools specifically for pediatric rheumatology disease populations; and (4) determine the barriers and facilitators to mental health screening in the pediatric rheumatology setting. There was general agreement between member subgroups regarding which research domains were of highest importance/feasibility/actionability. However, the domain of mental health screening had higher rankings among patients/parents and mental health providers than in the overall membership. Within each domain there was more often consensus than disagreement between subgroups about the ranking of research topics.

Small, single-center, cross-sectional studies have established that mental health symptoms are prevalent in youth with rheumatologic disease. However, our review of the literature uncovered that minority populations and diseases outside SLE and JIA were underrepresented. With so few studies, it is difficult to draw conclusions about the interaction between important socio-demographic factors or interactions with specific disease-related factors^{1,9}. Larger scale, multicenter studies will allow us to investigate sociodemographic risk factors and potential disparities in the identification of mental health symptoms across patient populations. Longitudinal studies are necessary to better understand the effect of disease-related factors and the nuances of mental health challenges across and within disease groups.

Topics related to mental health screening were rated most feasible and actionable and included 3 of the 4 highest priority topics. While mental health screening recommendations and guidelines exist for general pediatric populations^{17,18}, generalizing guidelines for primary care practitioners to rheumatologists is problematic. This is due to the increased prevalence of mental health conditions in youth with chronic conditions, the differing frequencies by which practitioners see patients, possible differences in mental health resources between pediatric subspecialty and general pediatric offices, and even differences in the relationships between patients and physicians among pediatric subspecialists and general pediatricians^{1,2,19}. Evidence supports systematic screening for mental health symptoms in adolescents with chronic illness²⁰. Screening approaches should also consider the known complex interplay between parental and child mental health and emotional adjustment to a chronic rheumatologic disease^{21,22,23}. Other chronic pediatric disease communities have developed specific screening tools^{24,25} and

screening and treatment guidelines around mental health symptoms for their populations¹¹. Implementation studies in models of pediatric chronic disease clinics show that mental health screening is both feasible and clinically meaningful^{10,20}. Yet specific guidelines regarding mental health screening of youth with rheumatologic diseases do not yet exist and are needed to address the unique needs of this population.

Mixed methods studies involving rheumatologists, mental health providers, patients and parents have identified anxiety, depression, emotional adjustment to illness, and caregiver emotional support as areas of high importance to be screened for and addressed in the pediatric rheumatology setting². Given the paucity of mental health resources specifically identified for this patient population, further screening studies should focus on these critical areas. Validation of screening tools is needed across disease subpopulations. Future studies will need to investigate implementation strategies in pediatric rheumatology clinical settings and the development and testing of interventions such as peer support groups and cognitive behavioral therapy to address mental health.

While initial work to define barriers and facilitators to mental health screening in pediatric rheumatology clinics has identified that a lack of resources, guidelines, and education stands in the way of broadly implementing screening practices, preliminary data in SLE show feasibility and patient/parent acceptability of screening for anxiety and depression in pediatric rheumatology clinics²⁶. This work needs to be broadened to other rheumatologic diseases, and implementation studies are needed to determine factors important to incorporating screening successfully into clinical practice. Quality improvement studies in pediatric rheumatology clinics are currently under way to optimize screening strategies to individual practices.

The research topic prioritization by workgroup members exposed research topics that are highly important and actionable but were not deemed highly feasible. Among these are topics concerning the burden and relationship of mental health disorders on rheumatologic disease and the effect of mental health on clinical outcomes. Ways to help improve the feasibility of researching these topics include the development of resources in pediatric rheumatology research to address questions around mental health. Enhancing capacity for multidisciplinary collaboration between rheumatology researchers and mental health professionals in psychiatry, psychology, and social work will bring the necessary mental health expertise into the context of pediatric rheumatologic disease. Expanding the current longitudinal CARRA disease registry to include instruments to measure mental health will also improve the feasibility of this type of research. Further, disease registries coupled to biorepositories and clinical data, such as the CARRA Registry, will help advance translational research to improve our understanding of pathophysiologic mechanisms that may help explain relationships between mental health symptoms and inflammation in rheumatologic disease.

There are several limitations to our study. First, despite inclusion of non-rheumatologists in the CARRA Mental Health Workgroup, the representation of mental health providers was minimal. This may be due to a lack of mental health providers connected to pediatric rheumatology clinics across the United States². Because their expertise is critical for multidisciplinary research to address mental health, further efforts are needed to recruit mental health professionals as integral members of research and care teams. Second, the representation of patients and parents was suboptimal. Their experiences with disease and as consumers of services provide essential perspectives for developing clinically meaningful and relevant research in mental health. Current and future projects of the workgroup aim for better representation and engagement of patients and families as key advisors and participants on the research team²⁷. Given the few members in both of these subgroups, we cannot infer whether differences in rankings reported in our study are

significant or truly representative. Third, the mental health research agenda is not representative of the entire CARRA membership. The prioritization process was limited to those involved in the Mental Health Workgroup. While this was an intentional design to gather input from those with knowledge and interest in mental health research, broader involvement of the CARRA membership will be necessary for successful collaborative projects including implementation studies of mental health guidelines in pediatric rheumatology.

Considering the challenges and limitations of investigating rare diseases such as those in pediatric rheumatology, leveraging the interest of multiple participants and identifying the most urgent and most feasible areas of research is imperative to foster successful and meaningful advances in research and care. By defining a prioritized research agenda to address the current gaps in knowledge that exist in our field regarding the mental health of youth with rheumatologic disease, we hope

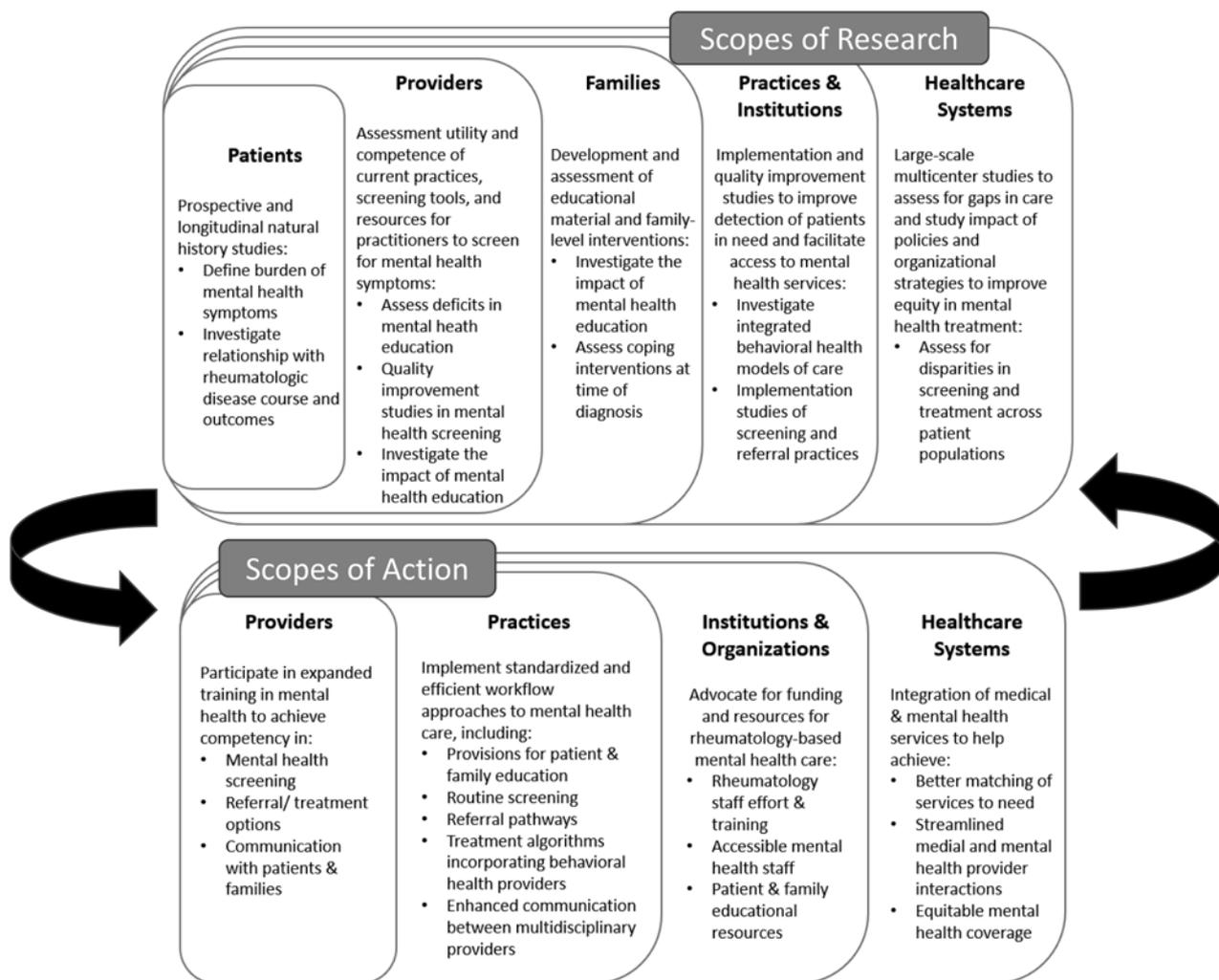


Figure 2. Responsiveness of CARRA Mental Health Workgroup prioritized research topics (Scopes of Research) to previously defined gaps in mental health care for patients with pediatric rheumatologic diseases (Scopes of Action)²⁸. The bidirectional arrows represent the iterative process between research and implementation activities to achieve improved care. CARRA: Childhood Arthritis and Rheumatology Research Alliance.

to improve care for this vulnerable patient population. These research topics address areas in need of improvement defined by our prior research²⁸ (Figure 2) and can direct specific research, clinical, and policy-making activities to improve care (Supplementary Table 1, available with the online version of this article).

The agenda developed here is intended to be an initial step in an iterative learning process requiring frequent reconsideration and revision. To improve upon the groundwork laid by the current agenda, we will need to expand partnerships with mental health experts and patient families, to develop revised future research agendas that better incorporate their knowledge and perspectives. Topics sufficiently researched will be checked off, making room for new topics that this agenda has yet to address. The success and the utility of the agenda in the future will be measured by whether the resulting studies lead to practice changes adopted by the larger pediatric rheumatology community and in further cycles of research to investigate whether those practices are producing improved outcomes for our patients.

ONLINE SUPPLEMENT

Supplementary material accompanies the online version of this article.

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