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Patient-Reported Outcomes Among Transition-Age Young Adults With Juvenile Idiopathic Arthritis in the Childhood Arthritis and Rheumatology Research Alliance Registry

Emily A. Smitherman[1], Rouba A. Chahine[2], Nicole Bitencourt[3], A.K.M. Fazlur Rahman[4], Erica F. Lawson[5], and Joyce C. Chang[6], for the CARRA Registry Investigators and CARRA Transition Workgroup

ABSTRACT. ***Objective.*** To evaluate patient-reported care utilization and outcomes among young adults with juvenile idiopathic arthritis (JIA), including factors associated with complete transfer to adult rheumatology.

Methods. We included young adults with JIA enrolled in the Childhood Arthritis and Rheumatology Research Alliance (CARRA) Registry from 2015 to 2019 with age ≥ 18 years at their last clinical site visit. We used data from the CARRA Registry Long-term Follow-up program, which follows inactive CARRA Registry patients and collects patient-reported information through phone surveys. We compared the characteristics of respondents with complete and incomplete transfer to adult rheumatology care at their first Long-term Follow-up phone survey.

Results. We identified 540 young adults with JIA; 187 (35%) responded to the Long-term Follow-up phone survey. The 54% of respondents with complete transfer to adult rheumatology were slightly older and reported more self-assessed disease activity, morning stiffness, and pain compared to those with incomplete transfer. Biologic use was high at both timepoints and did not differ by transfer status. Patients who completed the transfer were more likely to have private insurance and be actively pursuing postsecondary education compared to those with an incomplete transfer. Across the cohort, 65% reported problems with pain or discomfort and 45% with anxiety or depression.

Conclusion. Young adult respondents with JIA in the CARRA Registry commonly report persistent medication use, but still report more problems with pain as compared to population norms. Additional work is needed to understand how best to address comorbid pain around the period of transition to adult care.

Key Indexing Terms: juvenile idiopathic arthritis, outcomes, pain, quality of life, registries

The transition from pediatric to adult health care is a vulnerable period for young adults with childhood-onset rheumatic diseases, characterized by significant delays in routine follow-up, decreased treatment adherence, and increased emergency care utilization.[1-6] These gaps in care may lead to persistent disease activity and damage accrual.[2,7-12] Even with successful care transfer, the majority of patients experience transition difficulties, often related to loss of insurance or emotional adjustment.[8] Few studies have examined posttransfer psychosocial function and health-related quality of life (HRQOL) for young adults with childhood-onset rheumatic diseases. Studies of adults with juvenile idiopathic arthritis (JIA) have demonstrated higher educational attainment and employment rates compared to age-matched peers, as well as better HRQOL compared to patients with adult-onset arthritis.[13,14] However, studies of adults with childhood-onset systemic lupus erythematosus (cSLE) suggest poorer self-management skills, lower vocational attainment, and increased rates of anxiety and depression compared to those with disease onset in adulthood.[2,15,16]

Due to the complex developmental stage of adolescence and young adulthood exacerbated by the social vulnerability of transitions, there have been widespread efforts to systematically improve the process of pediatric to adult healthcare transition.[17] The Childhood Arthritis and Rheumatology Research Alliance (CARRA) Transition Workgroup has formed a transition learning collaborative to study the implementation of structured health care transition practices across multiple pediatric

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[1]E.A. Smitherman, MD, MS, Department of Pediatrics, University of Alabama at Birmingham, Birmingham, Alabama; [2]R.A. Chahine, PhD, Social, Statistical and Environmental Sciences Unit, RTI International, Research Triangle Park, North Carolina; [3]N. Bitencourt, MD, Department of Pediatrics, Loma Linda University Health, Loma Linda, California; [4]A.K.M.F. Rahman, PhD, Department of Biostatistics, University of Alabama at Birmingham, Birmingham, Alabama; [5]E.F. Lawson, MD, Department of Pediatrics, University of California San Francisco, San Francisco, California; [6]J.C. Chang, MD, MSCE, Department of Pediatrics, Harvard Medical School, Boston, Massachusetts, USA.

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Address correspondence to Dr. E.A. Smitherman, Department of Pediatrics, University of Alabama at Birmingham, 1600 7th Ave South, Birmingham, AL 35233, USA. Email: esmitherman@uabmc.edu.

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rheumatology centers.[18] However, a better understanding of current outcomes in young adults after leaving pediatric rheumatology care is also needed to study the effectiveness of transition processes and who benefits most from these strategies. To date, there have been few studies highlighting outcomes of the final phase of transition, following transfer to adult care.[19]

Our objective was to evaluate patient-reported care utilization, disease status, and HRQOL among young adults with childhood-onset rheumatic disease. We used data from the CARRA Registry Long-term Follow-up program that follows patients initially enrolled from pediatric rheumatology centers. We identified factors associated with complete or incomplete transfer to adult rheumatology.

METHODS

Data source. This study analyzed prospectively collected observational data from the CARRA Registry, which collects longitudinal data on patients across pediatric rheumatic diseases, including JIA, cSLE, and juvenile dermatomyositis (JDM).[20,21] Enrollment began in 2015, with over 70 pediatric rheumatology centers currently participating. Individuals participating in the CARRA Registry represent a convenience sample of patients treated at each site. Data in the CARRA Registry are collected within the context of routine clinical care every 6 (± 3) months. Written informed consent is obtained from all participants. This analysis was approved by the University of Alabama at Birmingham Institutional Review Board (IRB-170112004).

Patients are considered active at a CARRA Registry clinical site until they are referred elsewhere, move to a nonparticipating pediatric rheumatology center, transition to adult care, have no registry contact for more than 9 months, or attain disease remission with intended clinic visits less frequent than every 6 months. Established in 2016, the CARRA Registry Long-term Follow-up program allows for continued data collection from patients who are no longer active at a clinical site. After status is changed to inactive by the clinical site, the Long-term Follow-up program collects patient-reported information through phone surveys every 6 (± 3) months, with the goal to follow all patients for at least 10 years or until consent is withdrawn. Information is collected directly from patients who are aged ≥ 18 years. The program attempts to contact patients throughout the 6-month time window.

Study population. We analyzed data from patients enrolled in the CARRA Registry from 2015 to 2019 who had transitioned to the Long-term Follow-up program as of December 2019. All patients in the Long-term Follow-up program who were aged ≥ 18 years at the last CARRA Registry clinical site visit were eligible for inclusion. We limited our analysis to patients with JIA because of the small number of patients with cSLE (n = 25) and JDM (n = 1) meeting our criteria (Figure 1). We identified patients who responded to at least 1 Long-term Follow-up phone survey and used data from the first contact.

Measures. We extracted patient-level variables from the CARRA Registry: self-reported sex, race/ethnicity (combined as 1 variable within the registry), household income, highest parental educational attainment, insurance status at enrollment, primary rheumatic disease, International League Against Rheumatism (ILAR) category,[22] date of JIA diagnosis, and reason for inactive registry status. We extracted visit-level variables from the last recorded CARRA Registry clinical site visit: patient global assessment of well-being (PtGE) from 0 (very well) to 10 (very poor), physician global assessment of disease activity (0-10), number of active and limited joints, and morning stiffness. We extracted patient-reported variables from the first Long-term Follow-up contact: current age, current type of physician(s), transition preparedness ("how prepared did you feel in transferring your care?" on a 0-10 scale), hospital admission, emergency room utilization and if related to JIA, current steroid use, PtGE (0-10), patient-reported disease activity over the previous week (0-10), joint pain or swelling, morning stiffness, pain over the previous week (0-10), current insurance status, receipt of disability benefits or supplemental security income, highest year of school completed, employment status, financial status, marital status, current living situation (parent(s)/legal guardian, roommate, alone, significant other, other), and the 5-level EuroQol 5D (EQ-5D-5L).[23] The EQ-5D-5L

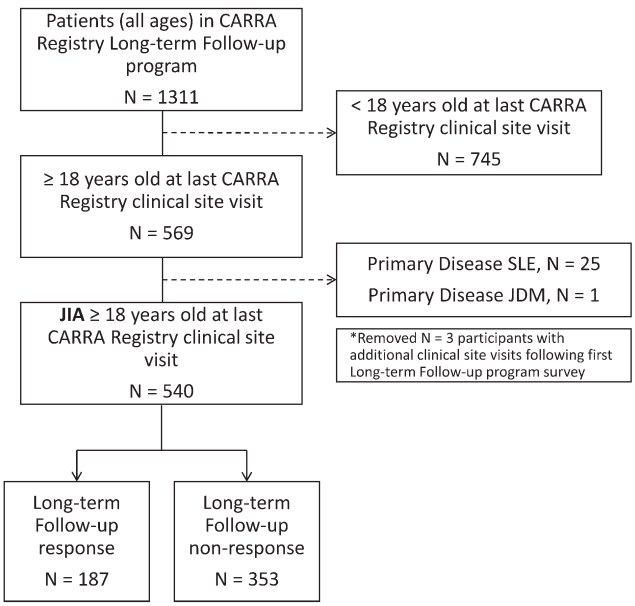

Figure 1. Selection process for patients included in the analysis. Of note, 3 participants had additional CARRA Registry clinical site visits following their first Long-term Follow-up survey and were removed from the analysis. CARRA: Childhood Arthritis and Rheumatology Research Alliance; JDM: juvenile dermatomyositis; JIA: juvenile idiopathic arthritis; SLE: systemic lupus erythematosus.

assesses the HRQOL domains of mobility, self-care, usual activities, pain/discomfort, and anxiety/depression on a 5-point Likert scale, along with a general health today visual analog scale (VAS), ranging from 0 (worst health imaginable) to 100 (best health imaginable). We extracted medications recorded as active at the time of the last clinical site visit and first phone survey. Medications were categorized as nonsteroidal anti-inflammatory drugs (NSAIDs); conventional disease-modifying antirheumatic drugs (cDMARDs) including azathioprine, cyclophosphamide, hydroxychloroquine, leflunomide, methotrexate, mycophenolate mofetil, tacrolimus, and thalidomide; or biologic DMARDs (bDMARDs) including abatacept, adalimumab, anakinra, canakinumab, certolizumab, etanercept, golimumab, infliximab, rilonacept, rituximab, secukinumab, tocilizumab, tofacitinib, and ustekinumab.

Statistical analysis. We compared sociodemographic and clinical characteristics of respondents and nonrespondents to the Long-term Follow-up phone survey. Among respondents, we grouped patients by complete transfer to adult rheumatology or incomplete transfer, defined as under the care of both an adult and a pediatric rheumatologist, a pediatric rheumatologist only, or no rheumatologist. We compared healthcare utilization, disease status, psychosocial functioning, and HRQOL between these groups using chi-square and Fisher exact tests for categorical variables and Wilcoxon rank-sum test for continuous/count variables. All tests were 2-sided with a significance level of 0.05 and conducted using SAS v9.4 (SAS Institute).

RESULTS

Characteristics of respondents vs nonrespondents to the Long-term Follow-up program. Of 1311 patients in the Long-term Follow-up program, 569 (43%) were aged ≥ 18 years at their last CARRA Registry site visit. Out of 540 young adults with JIA representing 57 North American pediatric rheumatology sites, 187 (35%) had responded to at least 1 Long-term Follow-up phone survey. The most common documented reasons for survey nonresponse were inability to contact the registry participant (82%) and the participant being unable to complete the phone interview at the time of contact (17%). A total of 8 participants withdrew from the study.

Nonrespondents were more likely to have lower reported household income (P = 0.03) and public insurance status (P = 0.03) at initial CARRA Registry enrollment compared to respondents (Table 1). The majority of respondents were White, and there was a tendency toward lower response rates in Black participants and those grouped in the other race and ethnicity category, though not in Hispanic participants. Disease duration at the last clinical site visit was on average 1.3 years longer among nonrespondents compared to respondents (P = 0.001), but no other differences in JIA disease status were identified. There was no difference in the documented reason for inactive registry status between the 2 groups.

Healthcare utilization. The 187 young adults with JIA with at least 1 Long-term Follow-up response had an average of 7.8 months between their last CARRA Registry site visit and first phone survey. Of these, 159 (85%) reported being under the care of any healthcare provider, but only 101 (54%) had a complete transfer to an adult rheumatologist. Of those with an incomplete transfer, 9 (5%) were under the care of both an adult and a pediatric rheumatologist, 21 (11%) reported remaining under the care of a pediatric rheumatologist only, and 56 (30%) were not under the care of any rheumatologist. A total of 97 (52%) of respondents reported receipt of primary care and 22 (12%) were receiving only primary care. Thirty-four (18%) also reported care from a nonrheumatology subspecialist.

"Transitioned to adult care" as determined by the CARRA site was the most common reason for inactive registry status (58%) across the cohort. However, 32% of those "transitioned to adult care" remained in the incomplete transfer group. There was also a higher percentage of patients categorized as "no registry contact > 9 months" in the incomplete transfer group (45% vs 18%). There was no difference in self-reported transition preparedness between those with complete transfer and incomplete transfer (median [IQR] was 9 [7-10] for both groups). At the time of the phone survey, 9% of respondents reported requiring hospital admission in the past 6 months, 5% reported emergency room visits, and 8% reported current steroid use with no differences between the 2 groups. Only 1 emergency room visit was reported to be related to JIA.

JIA disease status and medication use. The distribution of ILAR categories across the 187 young adults with JIA with Long-term Follow-up response was generally representative of JIA in the CARRA Registry (34% rheumatoid factor [RF]-negative polyarticular, 14% enthesitis-related arthritis, 14% RF-positive polyarticular, 13% oligoarticular, 11% psoriatic arthritis, 10% systemic arthritis, 4% undifferentiated arthritis).[21] During the phone survey, 7 respondents reported "don't know" when asked the name of their rheumatic diagnosis.

At the time of the first phone survey, young adults with complete transfer to adult rheumatology were on average 0.7 years older than those with incomplete transfer (P < 0.001) (Table 2). Those with complete transfer reported higher self-assessed disease activity (P = 0.01), more morning stiffness (P = 0.04), and higher pain scores (P = 0.03) compared to those with incomplete transfer. At the last CARRA Registry site visit, JIA disease activity measures tended to be low and were not different between respondents with and without a complete transfer. At the first phone survey, respondents tended to report worse global assessment of well-being, but lower rates of morning stiffness compared to their last clinical site visit, noting morning stiffness at the clinical site visit was unlikely to be strictly patient-reported.

There was no difference in active medications recorded at the last clinical site visit compared to the first phone survey (Figure 2). There was also no difference in medication categories at each timepoint when comparing patients with complete and incomplete transfer. The majority of patients reported bDMARD use as monotherapy or in combination with a cDMARD at both the last clinical site visit (60%) and the first phone survey (56%), of which 68% were a tumor necrosis factor (TNF) inhibitor. There was lower use of both cDMARD monotherapy and NSAID monotherapy. Approximately 30% of patients reported no active JIA medication use.

Psychosocial function and HRQOL. Overall, a majority of young adults with JIA reported favorable socioeconomic status including private insurance, completion of at least some college, full-time student status, and just enough money or some left over at the end of the month (Table 3). Patients who had complete transfer to adult rheumatology were more likely to be privately

Table 1. Characteristics of young adults with JIA in the CARRA Registry Long-term Follow-up program, by response group.

	Total Young Adults With JIA, n = 540	Long-term Follow-up Respondents, n = 187	Long-term Follow-up Nonrespondents, n = 353	*P*
Sociodemographics, n (%)				
Female sex	395 (73)	144 (77)	251 (71)	0.14
Race and ethnicity				0.06
White	393 (73)	147 (79)	246 (70)	
Hispanic	68 (13)	23 (12)	45 (13)	
Black	44 (8)	9 (5)	35 (10)	
Other[a]	35 (6)	8 (4)	27 (8)	
Household income, $				0.03
< 75,000	160 (30)	47 (25)	113 (32)	
≥ 75,000	173 (32)	73 (39)	100 (28)	
Unknown/prefer not to answer	207 (38)	67 (36)	140 (40)	
Highest parental education				0.72
High school or less	60 (11)	18 (10)	42 (12)	
At least some college	153 (28)	53 (28)	100 (28)	
Unknown/prefer not to answer	327 (61)	116 (62)	211 (60)	
Insurance status at enrollment				0.03
Private	385 (71)	145 (78)	240 (68)	
Nonprivate	148 (27)	39 (21)	109 (31)	
None	7 (1)	3 (2)	4 (1)	
JIA status at last site visit				
Time between diagnosis and last visit, mean (SD), yrs	7.7 (5.0)	6.8 (4.9)	8.1 (5.0)	0.001
PtGE (0-10), median (IQR)	1 (0-4)	1 (0-5)	1 (0-4)	0.62
PGA of disease activity (0-10), median (IQR)	0.5 (0-2)	0.5 (0-2)	0.5 (0-3)	0.49
Total no. of active joints, median (IQR)	0 (0-2)	0 (0-2)	0 (0-2)	0.79
Total no. of limited joints, median (IQR)	0 (0-1)	0 (0-1)	0 (0-1)	0.36
Presence of morning stiffness over the past week, n (%)	206 (38)	69 (37)	137 (39)	0.62
Reason for inactive registry status, n (%)				0.17
Transitioned to adult care	315 (58)	111 (59)	204 (58)	
No registry contact > 9 months	173 (32)	57 (30)	116 (33)	
Referred elsewhere	25 (5)	8 (4)	17 (5)	
Disease remission (clinic visits > 6 months)	17 (3)	4 (2)	13 (4)	
Subject moved to non-CARRA site	10 (2)	7 (4)	3 (1)	

P < 0.05 indicates significance. Wilcoxon rank-sum test was used to compare medians; chi-square or Fisher exact test was used to compare proportions. [a] Other includes Asian, Native Hawaiian/Pacific Islander, and Native American. CARRA: Childhood Arthritis and Rheumatology Research Alliance; JIA: juvenile idiopathic arthritis; PGA: physician global assessment; PtGE: patient global assessment of well-being.

insured ($P = 0.006$), to have completed some college education ($P = 0.003$), and to be a full-time or part-time student rather than employed ($P = 0.01$).

On the EQ-VAS general health today scale across the cohort, patients reported relatively good health with median (IQR) of 85 (70-90), and there was no difference based on transfer status. Patients reported almost no problems with self-care and some problems with usual activities and mobility (Figure 3). However, 65% reported problems with pain or discomfort (26% moderate to extreme), and 45% reported problems with anxiety or depression (27% moderate to extreme).

DISCUSSION

The results of this study illustrate short-term healthcare utilization, disease status, psychosocial function, and HRQOL in a group of young adults with JIA. With an average of 7.8 months between their last CARRA Registry visit at a pediatric rheumatology center and first follow-up phone survey, a little over half of young adults with an average age of 20 years reported complete transfer to an adult rheumatologist. Those with a complete transfer reported increased disease activity, including more morning stiffness and pain, compared to those with an incomplete transfer. Reported medication use did not change in this short-term period and did not differ based on transfer status. Participants with a complete transfer were slightly older, more likely to have private insurance, and more likely to have completed some college and be full- or part-time students. While social functioning and quality of life outcomes were generally positive, problems with pain and anxiety or depression were commonly reported. Our findings differ from previous studies across complex chronic conditions that found milder disease activity and older age at last pediatric visit were risk factors for gaps in care, whereas insurance status and educational attainment were not significant.[24]

Table 2. JIA disease activity at the first Long-term Follow-up phone survey compared to the last CARRA Registry clinical site visit in young adults with JIA, by transfer status.

	Total, n = 187	Complete Transfer to Adult Rheumatology, n = 101	Incomplete Transfer to Adult Rheumatology, n = 86	*P*
JIA status at first Long-term Follow-up phone survey (patient-reported)				
Average age at first survey, mean (SD), yrs	20.0 (1.4)	20.4 (1.4)	19.7 (1.2)	< 0.001
Time between last site visit and first survey, mean (SD), months	7.8 (4.0)	7.7 (4.2)	7.8 (3.9)	0.62
Time between diagnosis and first survey, mean (SD), yrs	7.5 (4.9)	7.8 (5.0)	7.0 (4.8)	0.23
PtGE (0-10), median (IQR)	2 (0-5)	3 (1-5)	1 (0-5)	0.39
Disease activity over the past week (0-10), median (IQR)	2 (0-5)	3 (1-5)	1.5 (0-3.5)	0.01
Any joint pain or swelling lasting 6 months, n (%)	111 (59)	62 (61)	49 (57)	0.58
Presence of morning stiffness over past week, n (%)	69 (37)	44 (44)	25 (29)	0.04
Pain over the past week (0-10), median (IQR)	3 (1-6)	4 (1-6)	2 (0-5)	0.03
Prior JIA status at last CARRA Registry clinical site visit (site-reported)				
PtGE(0-10), median (IQR)	1 (0-5)	2 (0-5)	1 (0-3)	0.15
PGA of disease activity (0-10), median (IQR)	0.5 (0-2)	1 (0-2)	0.5 (0-2)	0.33
Total no. of active joints, median (IQR)	0 (0-2)	0 (0-2)	0 (0-2)	0.85
Total no. of limited joints, median (IQR)	0 (0-1)	0 (0-2)	0 (0-1)	0.95
Presence of morning stiffness over past week, n (%)	100 (53)	58 (57)	42 (49)	0.36

Incomplete transfer category includes patients under the care of a pediatric rheumatologist only, both an adult and a pediatric rheumatologist, or no rheumatologist. $P < 0.05$ indicates significance. Wilcoxon rank-sum test was used to compare medians; chi-square or Fisher exact test was used to compare proportions. CARRA: Childhood Arthritis and Rheumatology Research Alliance; JIA: juvenile idiopathic arthritis; PGA: physician global assessment; PtGE: patient global assessment of well-being.

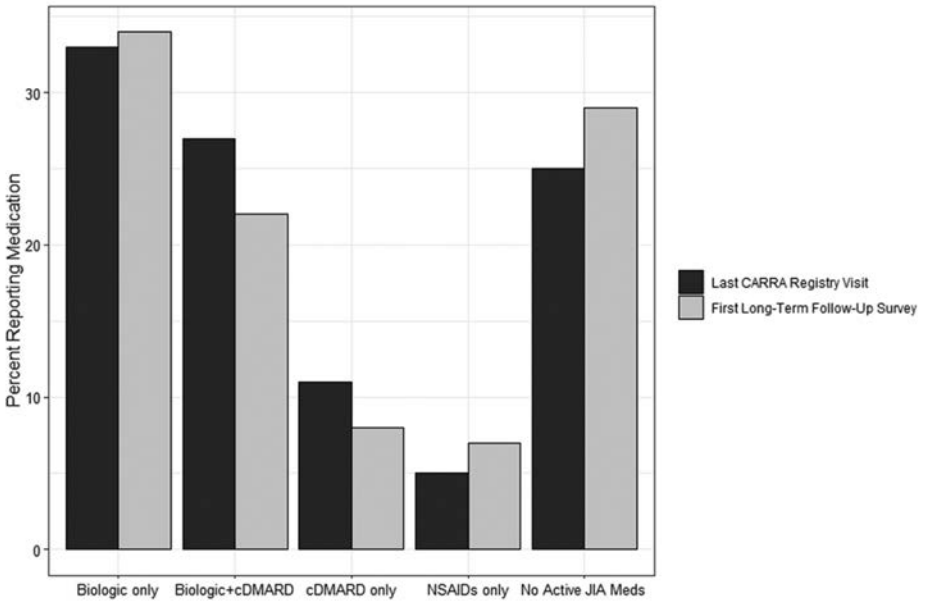

Figure 2. Active medication entries at last CARRA Registry clinical site visit and first Long-term Follow-up survey for young adults with JIA in the CARRA Registry Long-term Follow-up program (n = 187). Of patients reporting biologic medication use, 68% were a TNF inhibitor. CARRA: Childhood Arthritis and Rheumatology Research Alliance; cDMARD: conventional disease-modifying antirheumatic drug; JIA: juvenile idiopathic arthritis; NSAID: nonsteroidal anti-inflammatory drug; TNF: tumor necrosis factor.

Enrollment in the CARRA Registry began in 2015, and we examined data from patients with JIA enrolled through the end of 2019. Therefore, our study examines only shorter-term outcomes of young adults with an average JIA disease duration of 7.7 years, but with ongoing data collection, additional longitudinal studies will be possible over time. Further, since enrollment for cSLE began in 2017 and JDM began in 2018, additional data will soon be available to analyze for young adults with cSLE and JDM as more patients transition to the Long-term Follow-up program.

Our study is limited by a lower response rate within the Long-term Follow-up program, with data available for only 35% of eligible participants, although this is comparable to response rates of other national surveys. For example, the 2019

Table 3. Psychosocial function and HRQOL in patients with JIA ≥ 18 years at first Long-term Follow-up phone survey, by transfer status.

	Total, n = 187	Complete Transfer to Adult Rheumatology, n = 101	Incomplete Transfer to Adult Rheumatology, n = 86	*P*
Current private insurance status	148 (79)	88 (87)	63 (73)	0.006
Receiving disability benefits/SSI	11 (6)	6 (6)	5 (6)	0.99
Highest year of school completed				0.003
At least some college	115 (61)	72 (71)	43 (50)	
Grade 12 or GED[a]	60 (32)	22 (22)	38 (44)	
Other (Grade 9-11, prefer not to answer)	12 (6)	7 (7)	5 (6)	
Current employment status				0.01
Student	130 (70)	79 (78)	51 (59)	
Employed	36 (19)	12 (12)	24 (28)	
Other[b]	21 (11)	10 (10)	11 (13)	
Current financial status				0.43
Some left over at end of the month	38 (20)	24 (24)	14 (16)	
Just enough at end of the month	123 (66)	65 (64)	58 (67)	
Not enough at end of the month	23 (12)	11 (11)	12 (14)	
Never married	177 (95)	96 (95)	81 (94)	0.72
Current living situation				0.30
Parent/guardian	96 (51)	52 (51)	44 (51)	
Roommate	48 (26)	28 (28)	20 (23)	
Alone	22 (12)	12 (12)	10 (12)	
Significant other	14 (7)	8 (8)	6 (7)	
Other	7 (4)	1 (1)	6 (7)	
Average health today (0-100), median (IQR)	85 (70-90)	83 (70-90)	85 (75-90)	0.63

Values are expressed as n (%) unless indicated otherwise. Incomplete transfer category includes patients still under the care of a pediatric rheumatologist only, both an adult and a pediatric rheumatologist, or no rheumatologist. [a]GED is an alternative to a high school diploma in the US and Canada; most students commence grade 12 at 17-18 years of age, and GED candidates must be at least 16 years old, though often are 17 years old depending on location. [b] Other includes not employed, homemaker, disabled, and prefer not to answer. $P < 0.05$ indicates significance. Wilcoxon rank-sum test was used to compare medians; chi-square or Fisher exact test was used to compare proportions. GED: General Educational Development Test; HRQOL: health-related quality of life; JIA: juvenile idiopathic arthritis; SSI: supplemental security income.

National Survey of Children's Health had a 42% response rate from parents to mailed invitations,[25] and the 2007 Survey of Adult Transition and Health had a 17.5% response rate from young adults to telephone and internet invitations.[26] In order to understand potential nonresponse bias in our sample, we compared sociodemographics and JIA disease status at the last clinical site visit between Long-term Follow-up phone survey respondents and nonrespondents. We noted that respondents were more likely to have private insurance and a higher household income at initial CARRA Registry enrollment. This may potentially compound any existing selection bias at CARRA Registry enrollment due to differences between patients who are willing or not to provide informed consent. It may also decrease the generalizability of findings to all young adults with JIA in North America, particularly those from socioeconomically disadvantaged groups. A strategy employed by nationally conducted surveys that could be considered for future analyses involves calculating sampling weights to balance the differences found between respondents and nonrespondents.[27] Finally, the primary reason for nonresponse was the use of the telephone contact method, with patients either not answering the phone or unable to complete a questionnaire when reached. Response rates could potentially improve with asynchronous communication and data collection methods, such as text-messaging and online web-based surveys. Respondents to web-based surveys have been shown to have higher socioeconomic status compared to mailed surveys, so a multimodal approach may be most effective.[28] The CARRA Registry is actively implementing ongoing monitoring and feedback on accrual, eliciting patient feedback on approach strategies from those with concerns about participating, and employing multimodal strategies to contact participants to improve representative participation.

Our results demonstrated that only about half of patients aged ≥ 18 years with JIA had a complete transfer to an adult rheumatologist, which is largely consistent with previously published studies of pediatric to adult healthcare transfer outcomes.[5,6] This is in the context of high levels of perceived transition preparedness in both groups based on a 1-item question scaled 0 to 10 developed specifically for this registry. Although we were limited in our ability to more thoroughly assess transition preparedness because of the broad scope of the Long-term Follow-up phone survey, other posttransfer assessments have also observed high self-reported preparedness.[29] Even validated transition preparedness measures have been shown to not correlate with transition outcomes.[30] The high median and narrow range of scores we observed are likely a result of a ceiling effect with the question asked. A dedicated evaluation of transition preparedness in young adults with JIA is needed to more thoroughly understand its association with successful transfer, if any.

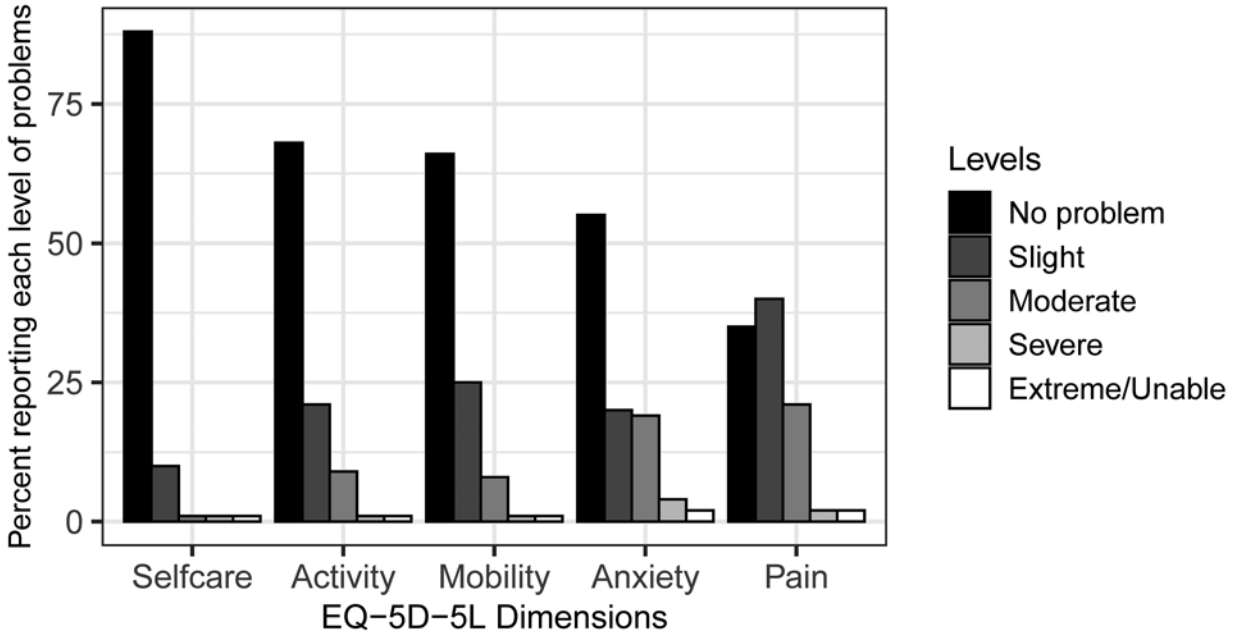

Figure 3. Reported problems in HRQOL (EQ-5D dimensions) among young adults with JIA in the CARRA Registry Long-term Follow-up program. CARRA: Childhood Arthritis and Rheumatology Research Alliance; HRQOL: health-related quality of life; JIA: juvenile idiopathic arthritis.

Almost 30% of phone survey respondents reported no active JIA medication use, similar to data from the last clinical site visit. While we are unable to discern missing medication entries in the site-reported data, missing entries are less likely given that the phone survey response was similar. Interestingly, our cohort also had high reported biologic use that persisted from the last clinical site visit (60%) to the first follow-up phone survey (56%), regardless of transfer status, a finding that is quite distinct from that of previously published studies. An analysis of a US private insurance claims database demonstrated a decrease in TNF inhibitor use among 58 youth with JIA from 33% while under pediatric care to 24% during the peritransfer interval and increasing again to 41% after establishing adult care.[1] A previous cohort of adolescent patients in Finland initiated bDMARDs mostly upon transfer to adult rheumatology.[12] Finally, an analysis of health plan claims from Germany demonstrated that only 56% of patients with JIA on DMARD therapy at 16 years of age remained on therapy at 20 years of age.[5] Although the higher rate of medication persistence in our sample may represent contemporary medication usage in North America, it may also reflect selection bias toward individuals with predominantly private health insurance and sufficient financial resources, or the short follow-up time period. Longer-term follow-up will be needed to determine whether medication persistence wanes over time.

Our study contributes to the growing literature on patient-reported outcomes (PROs) for young adults with JIA, including disease status, psychosocial function, and HRQOL within a short-term peritransfer period. Assessment of PROs allows for a more direct understanding of the patient's experience, behaviors, and health status.[31] The shift from largely clinician-reported data from clinical site visits to exclusively patient-reported data in the Long-term Follow-up program limits the comparison between these 2 data sources, although future longitudinal analyses of PROs can be conducted with ongoing data collection. Further, there may be risk of some recall bias regarding healthcare utilization over the 6-month period between Long-term Follow-up phone surveys. Contemporary short-term, posttransfer clinical data would also be beneficial to evaluate alongside PROs to better understand the drivers of patient perceived changes in disease status vs functional status. However, the ability to collect PROs directly from young adult patients during the peritransfer period provides an important opportunity to identify key elements to incorporate into transition interventions. In addition to programs focused on transition preparedness and improving the transfer process,[32-35] more detailed posttransfer outcomes data allow for a more targeted approach to programs supporting the needs of young adults with JIA, such as offering mental health resources for coping with pain.

In this cohort of young adults with JIA, we observed frequent reports of any problems with pain or discomfort and anxiety or depression in 65% and 45% of respondents, respectively. A 2017 US survey of people aged < 25 years reported any problems with pain or discomfort in 33% and with anxiety or depression in 42% of respondents using the same EQ-5D-5L scale.[36] Thus, reports of anxiety or depression in our cohort are only slightly higher than population norms, but reports of pain or discomfort are substantially higher. Additionally, the median pain scale is 3 (IQR 1-6) across the cohort and is significantly higher in young adults with a complete transfer to adult care compared to those with an incomplete transfer. Contemporary data on outcomes for young adults with JIA are lacking, but a recent description of a Nordic JIA cohort with an average age of 24 years documented a median (IQR) pain visual analog scale of 0.6 (IQR 0-3).[37] In our North American cohort, pain seems to be the predominant concern, despite established adult care, persistent medication use, and good overall health status. Additional work is needed

to better understand what accounts for this finding and how best to intervene, such as improving pain education and self-management support during transitional care.

In conclusion, young adults with JIA participating in the CARRA Registry Long-term Follow-up program report good transition preparedness, persistence of JIA-related medication use, and positive social functioning. We identified a need to improve the number of patients who complete transfer to adult rheumatology and to better understand the etiology and impact of pain in young adults with JIA. These initial results can inform the ongoing work of initiatives, such as the CARRA Transition Learning Collaborative, to identify the most effective peritransfer interventions to optimize the long-term health outcomes among people with JIA. This includes improving access to adult providers and patient support programs. There is also a continued need to develop better strategies for evaluating patient-reported disease status and HRQOL for young adults most at risk for disparate outcomes who may not be represented in the current cohort.

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