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Building a learning health system in pediatric rheumatology

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Building a learning health system in pediatric rheumatology

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Editorial: Building a learning health system in pediatric rheumatology

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Editorial on the Research Topic
Building a learning health system in pediatric rheumatology

Pediatric rheumatic diseases are rare, immune-mediated illnesses that often have a chronic course. Affected children may face a lifetime of challenges including potential disability, persistent pain, and the long-term impact of medications and medical interventions. Early diagnosis and prompt treatment by pediatric rheumatologists using shared decision making can reduce future morbidity. However, many children lack access to timely specialty care due in part to a critical pediatric rheumatology provider workforce shortage, and also limited societal awareness of autoimmune diseases in children resulting in delayed recognition and referral. This vulnerable population of children is left underserved and with their health outcomes uncertain.

In this context, the learning health system (LHS) model offers a promising framework for improving healthcare delivery in pediatric rheumatology (1). A LHS integrates clinical data with continuous quality improvement (QI) and implementation science to enhance outcomes, generate knowledge, and foster research. A community focused on outcomes improvement and a culture of active knowledge sharing is foundational to the success of a LHS. With an estimated 300,000 children in the U.S. affected by rheumatic diseases (2), access to care remains inequitable. The affected population is dispersed across urban and rural settings and is served by pediatric specialists employed at select academic medical centers nationwide, where cognitive subspecialties caring for children are afforded few financial resources. In rare diseases, a shared clinical registry is essential to aggregate sparse data, conduct meaningful analyses, and drive improvements. The LHS model's ability to address health care delivery gaps, drive outcomes improvement, and advance clinical care and knowledge makes it particularly relevant in pediatric rheumatology.

To explore the impact of LHS in this field, we put out a call for a special Research Topic on "Building a Learning Health System in Pediatric Rheumatology". Our goal was to highlight real-world clinical practice and network interventions to improve the quality of healthcare delivery and outcomes in pediatric specialty care. The editorial

team is pleased to highlight a broad overview of the 12 articles selected by peer review for publication, representing 23 distinct health systems across North America.

Key themes reflecting drivers of improving chronic illness care emerged across these contributions. A study by Vora et al. addressed delays in referral and low access to care. The authors describe a single center intervention to increase timely referrals to rheumatology care from safety net primary care clinics through education, raising awareness of presenting symptom bundles of rheumatic diseases, streamlining referral processes, and establishing a triage system with expedited scheduling for urgent cases. This work exemplifies the capability of a single clinic to influence access and timeliness of care within the confines of a larger health system.

Another major theme was addressing variation in treatment practices. Balay-Dustrude et al. describes unwarranted variation in care in a then nascent learning health network (Pediatric Rheumatology Care and Outcomes Improvement Netowrk, PR-COIN), through evaluation of use of intra-articular corticosteroid injections (IACI) following release of American College of Rheumatology JIA treatment guidelines (in 2011, 2013). Data from 2011 to 2015 was analyzed on whether IACI were used as primary treatment for oligoarticular JIA. Although there was no network-level intervention, centers worked locally to try and achieve a network stated goal of IACI within 2 weeks of diagnosis. Despite local efforts, results revealed lower-than-expected IACI rates and regional practice variation, highlighting the need for standardized care approaches within a LHS.

Multiple articles touched on the importance of data capture to assess patient outcomes to evaluate for potential gaps in care or disparities, and of the reliable collection of disease specific activity measures across patient population. Goh et al., and Pan et al., conducted surveys of PR-COIN LHS members to understand current state of data collection across network centers, and understand barriers and facilitators to reliable collection. Goh focuses on the role of 'critical data elements' and offers unique insights on the challenge of collection during tele-rheumatology visits. Patient centered care requires assessment of outcomes directly from the patients (PROs), Pan shares best practices for systematic PRO collection based on network experience. Although health equity is a key principle of quality of care, a fundamental prerequisite is data completeness which confers the ability to identify disparities. Banschbach et al., through an evaluation of PR-COIN learning network registry data identified missing demographic data in 1/3 of patients. With interventions, this was remediated in about 94%. The article noted that patients missing race data are likely to be missing other critical data elements, and challenges us to equitably measure disease activity and ensure data capture for vulnerable groups.

Two studies detailed single-center interventions leveraging electronic health records (EHR) for data-driven improvement. Timmerman et al. and Barbar-Smiley et al., each provide single center interventions to standardize disease activity data collection in the electronic health record (EHR). Timmerman describes partnership with local IT resources to establish a center dashboard to capture data on all clinic patients to support QI initiatives. Barbar-Smiley's group leveraged features within the EHR collection system to support and sustain a clinic effort to capture systemic lupus (SLE) disease activity, a process that also benefitted their research registry participation.

Huang et al. describe use of technology to leverage clinical registry data of an LHS to refine algorithms supporting selection of personalized treatment based on real world experience of other similar patients, which can be used at point of care in shared decision making with patients.

Mental health and self-management support were also focal points. Harper et al. present an intervention to systematically collect mental health data in childhood SLE patients, and an intervention to respond with mental health supports. Argraves et al., address the perpetual challenge of assessing and supporting transition readiness in children reaching age of majority. They leverage process automation to achieve reliability in assessment (90%). The intervention also requires provider review and self-management support education. That multiple QI interventions were published from one center suggests a strong culture of QI, and effective use of QI as a strategy for operational improvement.

Two articles highlighted outcomes improvement and parent engagement within PR-COIN, a pediatric rheumatology LHS with over 7,200 active patients across 23 centers. Harris et al. and Ferraro et al., discuss network-wide interventions and outcome improvements demonstrating the effectiveness of the LHS model in driving sustainable improvements. Of vital importance to the network's success is the engagement of parents and patients in design and conduct of interventions contributing the patient perspective, and keeping focus on patient outcomes.

What can we take away from this issue? The inspiration of pediatric rheumatology clinical teams working in coordination together with parents to move the needle on healthcare improvement for a vulnerable patient population. A culture of QI, rigorous data collection, and the use of technology are central to this work. By focusing on equitable access, timely diagnosis and treatment, mental health integration, proactive care teams, engaged families, and health system support for QI, a LHS organizational framework has been shown to improve processes and patient outcomes in pediatric rheumatology. This special issue serves as a testament to the power of adopting a LHS model in transforming care in pediatric chronic illness care.

Author contributions

EM: Writing – original draft, Writing – review & editing. CM: Writing – review & editing. SV: Writing – review & editing. BG: Writing – review & editing.

Conflict of interest

Seattle Children's Research Institute is the coordinating center for Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN). EM is employed by Seattle Children's and serves as the principal investigator of PR-COIN. EM, BG, SV serve in volunteer capacity on steering committee of PR-COIN.

The remaining author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Generative AI statement

The author(s) declare that no Generative AI was used in the creation of this manuscript.

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Intra-articular corticosteroid utilization and characterizations of use in juvenile idiopathic arthritis within the PR-COIN registry

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Objective: Intra-articular corticosteroid injections (IACI) have been shown to be effective at improving arthritis across juvenile idiopathic arthritis (JIA) categories. The American College of Rheumatology (ACR) recommends IACI use as primary and adjunctive therapy for JIA patients. However, there remains minimal data describing actual IACI use in North America. The objective of this study was to describe and to evaluate IACI use in JIA, utilizing the Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) registry.
Methods: Study participants from 13 sites were enrolled in the PR-COIN registry from 2011 to 2015. Demographic and clinical variables were summarized and Chi-squared and *t*-tests were used to evaluate differences between participants who did or did not receive IACI. Multiple logistic regression models were used to evaluate characteristics associated with IACI treatment.
Results: Our study included 3,241 participants, the majority of whom were white (85%), female (71%) and had oligoarticular JIA (39%). IACI was administered at least once in 23% of participants, the majority of whom had oligoarticular disease (52.5%), but overall use in oligoarticular participants was low at 30.8%. IACI use varied significantly between treatment centers and use was associated with oligoarticular disease, ANA positivity, and use of other systemic medications.
Conclusion: This study demonstrates that participants with JIA enrolled in the PR-COIN registry between 2011 and 2015 with persistent oligoarticular disease, ANA positivity, and use of other systemic medications were more likely to receive IACI. However, IACI use was lower than expected for oligoarticular participants.

KEYWORDS

juvenile idiopathic arthritis, corticosteroid injection, treatment, outcome, registry

1 Introduction

Juvenile idiopathic arthritis (JIA) is one of the most common chronic rheumatic diseases of childhood, with a prevalence of approximately 1 per 1,000 population (1). The International League of Associations for Rheumatology (ILAR) classification includes seven JIA subtypes. The most common subtype, oligoarticular disease (≤4

joints involved), accounts for 50%–80% of all chronic arthritis cases in North America and Europe (1). Expeditious and effective treatment of JIA is required in order to relieve pain, promote growth, and prevent permanent functional disabilities and joint destruction (2, 3).

A variety of treatment modalities are available for use by the pediatric rheumatologist to arrest the inflammatory process and achieve disease control. The 2011, 2013, 2019 and 2021 American College of Rheumatology (ACR) JIA treatment guidelines have consistently recommended intra-articular corticosteroid injections (IACI) as a primary treatment for oligoarticular disease and as adjunct or bridging therapy for polyarticular disease, sacroiliitis, and systemic disease. IACI are often used in combination with other therapeutics including non-steroidal anti-inflammatory drugs (NSAIDs), conventional synthetic disease modifying antirheumatic drugs (csDMARDs), and biologic DMARDs (2–5). The variety of treatment options and lack of evidence to specifically recommend one treatment over another, which is highlighted by the ACR guidelines, has resulted in varying strengths of practice recommendations, and likely varying levels of adherence to these recommendations.

Evidence of the effectiveness of IACI in the treatment of JIA is based on a number of retrospective and prospective studies which define effectiveness as a prolonged period of inflammatory inactivity in the injected joint after treatment (6–10). However, achievement of disease inactivity and the duration of efficacy after IACI varies depending on JIA subtype, age, disease duration, antinuclear antibody (ANA) positivity status, concomitant systemic therapy, and preparation of intra-articular glucocorticoid used (7, 10–14). Furthermore, ACR guidelines and multiple studies demonstrate favor triamcinolone hexacetonide (TH) over triamcinolone acetonide (TA), as it has been shown to induce longer periods of remission in injected joints (4, 5, 9, 15–17). In two fairly recent studies, the response rate to IACI, which was defined as absence of arthritis at 6 months, ranged from~50%–70% (9, 10). It would be expected, then, that many children require other therapies beyond IACI.

While these studies provide a foundation for treatment, and the initial 2011 ACR guidelines recommended IACI use, there is limited data on the actual clinical context in which IACI are utilized and the prevalence of use in a large population of JIA patients. Given this knowledge gap, the goal of this study was to evaluate the baseline use of IACI in the treatment of JIA in a large North American (United States and Canada) cohort. We examined the prevalence and predictors of IACI use in participants with JIA who were enrolled in the Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) registry between January 1, 2011, and July 31, 2015.

PR-COIN is a multicenter "Learning Network" in North America that uses quality improvement methods to develop and evaluate JIA management strategies, with a goal of improving disease outcomes for children with JIA. The network focuses on collection of data that can be used at the point of care to inform treatment decisions, with an emphasis on close partnership with patients and their families. Implementation of interventions proven to improve chronic illness care – for example pre-visit planning, population management, and shared decision making – and how such interventions impact disease outcomes is a priority (18). While PR-COIN has an established goal to administer IACI in a timely fashion, within 2 weeks of identified need, there has not yet been a network-wide initiative around the use of this treatment modality. This study, then, serves to shed light on baseline treatment patterns within the network. It helps identify known but important gaps in data collection and helps frame the need for future work on how treatment decisions ultimately impact outcomes.

2 Methods

2.1 Population

De-identified data for all children with JIA enrolled in the PR-COIN registry from 2011 inception through July 2015 were extracted and analyzed. Any patient with JIA at a participating PR-COIN center was eligible for enrollment in the registry though the actual process of enrollment varies by center. For example, some centers require patient consent to enroll while others do not. Notably, registry enrollment occurred between 2011 and July 2015 though participants may have been diagnosed or received treatments prior to their enrollment. Treatment information reported reflects treatments administered during this time frame from 2011 to 2015, or prior to enrollment when reported. Because data was de-identified, analysis and results could not include specific dates as only days/months from an unknown referent date were provided.

This project was approved by the Institutional Review Board (IRB) and not considered human subjects research. Data included patient demographics, ILAR subtype, diagnosis and encounter dates (listed as days from referent point), ANA positivity status, and treatments. Treatments captured included: any NSAID, IACI (triamcinolone acetonide, triamcinolone hexacetonide, and other), non-biologic DMARDs (azathioprine, cyclosporine, hydroxychloroquine, leflunomide, methotrexate, sulfasalazine, and other) and biologics (abatacept, adalimumab, anakinra, canakinumab, certolizumab, etanercept, golimumab, infliximab, rilonacept, rituximab, tocilizumab, and other). Data from participants enrolled at 12 out of 13 eligible medical centers were analyzed; one center did not have IACI data available and was therefore excluded. For some participants who had discrepant diagnosis dates, the earliest reported date was used.

2.2 Statistical analysis

Demographic and clinical factors were analyzed for differences between those who received at least one IACI and those who did not receive any IACI using Chi-square tests and *t*-tests for categorical and continuous measures, respectively. Multiple logistic regression models, along with odds ratios and 95% confidence intervals, were used to investigate demographic or disease features associated with the odds of receiving IACI. Site, age at diagnosis, ANA status, ILAR code, race, ethnicity, NSAID,

DMARD, and biologic DMARD use were selected *a priori* as relevant factors to be included in the model. Some participants received more than one IACI, so analysis was run using both minimum and maximum assumptions for administration, meaning that when there was a report of IACI use in the data, but no date was listed, the minimum assumption was that the missing date was one of the subject's other non-missing date values. The maximum assumption was that each missing date was assumed to be a unique date. As these results were without significant differences, the minimum IACI use assumption results were reported. When there were missing, unknown, or incomplete data, these were excluded from the regression model. R (R Core Team) Version 4.0 was used for all analyses. *P*-values less than 0.05 were considered statistically significant.

3 Results

3.1 Registry data and population demographics

There were 3,241 participants enrolled in the PRCOIN registry from 2011 to 2015 and included in the analysis, with 14 participants excluded for incorrect or missing data. The majority of registry participants were from the United States (81.5% vs. 18.5% from Canada), White (85%), female (71%), and had oligoarticular disease (39%, persistent and extended), similar to previously reported North American characteristics of participants with JIA (Table 1) 0/0/00 0:00:00 AM (19, 20). Persistent oligoarticular participants were defined as those with ≤4 joints involved, while extended oligoarticular participants were those whose joint count extended to involvement of >4 joints after the first 6 months of their disease. There was some variability of ILAR subtype by site (Supplementary Table S1). Patient duration in the registry varied, with 23.6% (765/3,241) having only one visit recorded, 29.5% (956/3,241) enrolled in the registry for less than a year, and 46.9% (1,519/3,241) enrolled for a year or more. Time from diagnosis to first registry encounter was 48.4, 48 months (mean, SD), meaning many patients were enrolled well into their diagnosis.

TABLE 1 General demographic and disease/treatment features of PR-COIN registry participants from 2011 to 2015.

Characteristic	*N* = 3,241*
Age at diagnosis	
Mean age in years (SD)	7.6 (4.8)
Months between diagnosis and 1st registry visit	
Mean duration in months (SD)	48.4 (48.0)
Duration in registry	765 (23.6%)
1 visit	956 (29.5%)
<1 year	1,519 (46.9%)
≥1 year	
Gender	
Female	1,937 (70.9%)
Male	795 (29.1%)
Race	
Non-White	388 (15.3%)
White	2,149 (84.7%)
Ethnicity	
Hispanic	291 (11.7%)
Non-Hispanic	2,192 (88.3%)
ILAR Code	
Oligoarticular persistent	993 (30.7%)
Oligoarticular extended	276 (8.5%)
Polyarticular RF (+)	172 (5.3%)
Polyarticular RF (-)	883 (27.3%)
Psoriatic arthritis	219 (6.8%)
Enthesitis related	398 (12.3%)
Systemic	208 (6.4%)
Undifferentiated	84 (2.6%)
Country	
US	599 (18.5%)
Canada	2,641 (81.5%)
IACI use^	
TH	513 (15.8%)
TA	300 (9.3%)
Other	23 (0.7%)
None	2,494 (77%)
Any IACI use	747 (23%)
Any NSAID use	2,049 (63.2%)
Any DMARD use	1,739 (53.7%)
Any biologic use	1,495 (46.1%)

SD, standard deviation; RF, rheumatoid factor; US, United States; IACI, intra-articular corticosteroid injection; TH, triamcinolone hexacetonide; TA, triamcinolone acetonide; ILAR, international league against rheumatism; IACI, intra-articular corticosteroid injection; NSAID, non-steroidal anti-inflammatory drug; DMARD, disease modifying anti-rheumatic drug.
*Some values may not add up to total due to missing data.
^Some patients received more than one type of IACI.

3.2 IACI Use

Twenty-three percent (747/3,241) of participants received one or more IACI, with TH being the corticosteroid formulation used most often, accounting for 61.3% of IACIs given (513/836). The median time to first captured injection for those who received IACI treatment was 28 months (range 25–230 months) after diagnosis. Retrospective treatment data was not required for registry enrollment so there are likely instances of IACI prior to what was captured. Negative values reflect IACIs received before participants were enrolled in the registry but after their initial diagnosis. IACI was the least commonly used treatment, with other treatments (NSAIDs, DMARDs, and biologics) being used in a greater percentage of participants overall (Table 1), keeping in mind that individual participants may receive multiple therapies either in sequence or concurrently. Data for sequence of medication, or multiple medications administered within the same class, for example repeat IACI use, was limited and incomplete so was not included in this analysis.

The majority of IACI were administered in participants who had oligoarticular disease, which accounted for 52.5% (391/745) of the IACI use (Table 2). However, considering that IACI is a main treatment modality recommended in this subtype, then the overall use in participants with oligoarticular disease remained low, with only 30.8% (391/1,269) of participants with oligoarticular disease receiving IACI. This indicates that 69.2%

TABLE 2 Comparison of features for patients who did vs. did not receive intra-articular corticosteroid injections.

Characteristic	IACI use - yes (*N* = 747^)	IACI use - no (*N* = 2,494^)	*P*-value
Center**			<0.001*
Country			0.525
United States	614 (82.3%)	2,027 (81.3%)	
Canada	132 (17.7%)	467 (18.7%)	
Region			<0.001*
Canada (2 sites)	132 (17.%)	467 (18.7%)	
US Midwest (2 sites)	206 (27.6%)	452 (18.1%)	
US Northeast (4 sites)	301 (40.3%	1,035 (41.5%)	
US South (4 sites)	77 (10.3%)	270 (10.8%)	
US West (4 sites)	30 (4.0%)	270 (10.8%)	
Duration in registry			<0.001*
One visit	64 (8.6%)	701 (28.1%)	
<1 year	186 (24.9%)	770 (30.9%)	
≥1 year	497 (66.5%)	1,022 (46.9%)	
Age at diagnosis (years)			0.008*
Mean (SD)	7.1 (4.7)	7.7 (4.9)	
Range	0.7–17.9	0.2–23.7	
Gender			0.012*
Female	469 (74.9%)	1,468 (69.7%)	
Male	157 (25.1%)	638 (30.3%)	
Race			0.004*
Non-White	69 (11.6%)	319 (16.4%)	
White	525 (88.4%)	1,624 (83.6%)	
Ethnicity			0.165
Hispanic	58 (10.1%)	233 (12.2%)	
Non-Hispanic	517 (89.9%)	1,675 (87.8%)	
ILAR Code			<0.001*
Oligoarticular persistent	311 (41.7%)	682 (27.4%)	
Oligoarticular extended	80 (10.7%)	196 (7.9%)	
Polyarticular RF (+)	33 (4.4%)	139 (5.6%)	
Polyarticular RF (-)	195 (26.2%)	688 (27.7%)	
Psoriatic	41 (5.5%)	178 (7.2%)	
Enthesitis related	52 (7.0%)	346 (13.9%)	
Systemic	22 (3.0%)	186 (7.5%)	
Undifferentiated	11 (1.5%)	73 (2.9%)	
ANA status			<0.001*
Positive	439 (58.8%)	1,104 (44.3%)	
Negative	281 (37.6%)	1,207 (48.4%)	
Unknown/missing	27 (3.6%)	183 (7.3%)	
Any NSAID use	594 (79.5%)	1,455 (58.3%)	<0.001*
Any DMARD use	465 (62.2%)	1,274 (51.1%)	<0.001*
Any biologic use	355 (47.5%)	1,140 (45.7%)	0.383

SD, standard deviation; RF, rheumatoid factor; US, United States. IACI, intra-articular corticosteroid injection; ILAR, international league against rheumatism; ANA, antinuclear antibody; NSAID, non-steroidal anti-inflammatory drug; DMARD, disease modifying anti-rheumatic drug.

^Individual categories may not add up to this number due to missing data.

*Statistically significant difference for characteristic.

**Center specific IACI use available in Supplementary Table S2.

(878/1,269) either did not receive IACI therapy or that this was not captured. For participants with polyarticular disease (rheumatoid factor positive and negative), 21.6% (228/1,055) received IACI, followed by 18.7% (41/219), 13% (42/398), 10.5% (22/208), and 13% (11/84) for patients with psoriatic arthritis, enthesitis related arthritis, systemic JIA, and undifferentiated disease respectively.

There were statistically significant differences among those who did vs. did not receive IACI by center, indicating that treatment practices may vary among clinical sites (Table 2). For example, less than 10% (5/51) of those at site B received an IACI compared to just over 30% (145/475) at site A (Supplementary Table S2). There was no statistically significant difference between sites based on country (United States vs. Canada). When centers were grouped by region, there was a detectable difference, with centers in the United States Midwest demonstrating the highest rate of IACI use (206/658 = 31.3%) and US western centers demonstrating the lowest use (30/300 = 10.0%) (Table 2).

There were also notable differences for IACI use based on duration in the registry. Participants with only one visit recorded in the registry (64/765 = 8.4%) or less than 1 year of registry participation (186/956 = 19.5%) demonstrated lower IACI use compared to participants in the registry for a year or more (497/1,519 = 32.7%) (Table 2).

3.3 Characteristics of IACI recipients

There were statistically significant differences for IACI use by patient ILAR subtype, gender, race, and ANA status (Table 2). The use of NSAIDs and DMARDs was greater among those who received IACI as compared to those who did not, while biologic use between the groups was similar.

Similarly, when accounting for these various characteristics through a logistic regression model, we found that treatment center, ILAR category, ANA positivity, and use of other systemic medications were associated with greater odds of receiving IACI (Table 3). Participants with persistent oligoarticular disease expectedly had the highest odds of receiving IACI treatment, when adjusting for all the other factors in the model.

4 Discussion

The goals of treatment for patients with JIA encompass the use of safe, timely, and effective medication to achieve disease control. Ultimately, understanding which therapies are most effective for which patients will inform medical decision-making and lead to improved clinical outcomes. Achieving this goal requires rigorous comparative effectiveness studies, designed in an informed, data driven manner. This study, a secondary analysis of existing data from the PR-COIN registry, is a first step in understanding how IACI are being used in clinical practice and whether there are predictors of their use. This, in turn, can help shed light on what other factors that may contribute to use should be captured and evaluated in future studies.

In this study, we observed that prevalence of IACI use varied by treatment center. When comparing centers in the United States vs. Canada, we did not observe a difference in use; however, when divided further to include regions within the United States, there were observed differences. This may indicate that therapy decisions are driven less by insurance coverage or availability of medication, for example, as these would likely differ more between countries. Rather, perhaps there are factors driven by local culture,

TABLE 3 Logistic regression *p*-values and odds ratios for significant comparisons.

Parameter	Odds ratio	95% confidence interval	*p*-value
Center^			<0.001*
Age at diagnosis			0.20
Gender			0.57
Race			0.29
Ethnicity			0.54
ANA status			0.011*
Positive vs. negative	1.31	1.06–1.63	
ILAR code			<0.001*
Oligoarticular persistent	ref	–	
Oligoarticular extended	0.64	0.43–0.93	
RF + polyarticular	0.36	0.20–0.57	
RF - polyarticular	0.41	0.31–0.54	
Psoriatic	0.36	0.21–0.53	
Enthesitis-related	0.24	0.15–0.35	
Systemic	0.20	0.14–0.44	
Undifferentiated	0.24	0.09–0.52	
NSAID use			<0.001*
Yes vs. no	2.61	2.03–3.39	
DMARD use			0.002*
Yes vs. no	1.35	1.08–1.93	
Biologic use			<0.001*
Yes vs. no	1.52	1.20–1.93	

Reference group use for ILAR code associations: Persistent Oligoarticular.
RF, rheumatoid factor; ANA, antinuclear antibody; ILAR, international league against rheumatism; NSAID, non-steroidal anti-inflammatory drug; DMARD, disease modifying anti-rheumatic drug.
^Specific comparisons and odds ratios not included here due to multiple possible pairwise comparisons.
*Statistically significant comparison.

number of providers at a center, availability of ancillary services such as pediatric anesthesia and child life specialists, and whether the hospital is a teaching/training institution.

Our cohort, which includes prevalent cases of JIA enrolled in the PR-COIN registry between 2011 and 2015, captures a time period of IACI use prior to and during the early stages of the ACR guidelines for IACI use. Given the mean time from disease diagnosis to registry enrollment (48.4 months), treatments administered shortly after diagnosis, which may well have been IACI use, were often not captured in the registry with enough detail to evaluate in this analysis. Thus, it is not unexpected that we observed IACI use to be overall lower than recommended by the ACR guidelines, particularly in those with oligoarticular disease. We also found that other medications including NSAIDs, biologics and conventional DMARDs were associated with treatment with IACI, which we interpret to mean that within this cohort, IACI were not the primary treatment modality in most cases but rather an adjunct therapy. This may indicate that IACI do not necessarily alleviate the need for additional rheumatologic treatments to achieve disease control but are used often as bridging or adjunct therapy, a concept that is supported by other studies (9, 10). This consideration is further corroborated by Papadopouluo et al., who demonstrated that IACI may be utilized as bridge therapy while awaiting systemic therapy to take effect and have been shown to be effective even in polyarticular patients with the benefit of avoiding or limiting systemic corticosteroid therapy and its side effects (21).

Regarding selection of injectable corticosteroid medication, in the PR-COIN registry, TH was used at a higher rate than TA. This finding likely indicates a preference for this medication when available, as our data represents a time period when TH was still widely available in the US, prior to its discontinuation in 2015. These findings are in line with the available literature, both historical and recent, which shows a clear benefit in terms of longevity of efficacy for IACI with TH over TA (9, 17, 22). Given the clinical superiority of TH, one might question why it did not account for an even greater percent of IACI in our cohort.

Further, we sought to understand which participants received IACI most frequently in the PR-COIN registry. In this study, we observed differences in recipients of IACI by ILAR code, gender, race and ANA status. This matches what might be expected, with use being higher for oligoarticular disease, more common among females and those who are white, which accounts for most patients with JIA and in particular those with oligoarticular disease, and among those who are ANA positive, with ANA positivity being most common in the oligoarticular subtype. Using a logistic regression model, we identified potential predictors of IACI use which included: treatment center, ILAR category, and ANA positivity. This similarly aligns with what we would expect.

Finally, the majority of the available literature surrounding IACI use focuses on predictors of disease course after injection. However, there has been limited evaluation of predictive factors for the use of IACI as a first line of treatment in JIA, despite evidence to suggest that IACI were most effective for young JIA patients with a short disease course (20). Thus, further evaluation of which patients are most likely to both receive and benefit from IACI early in the disease course is warranted. This gap in the literature, in combination with our study results, suggest there may be under-utilization of IACI in this population of JIA patients, and advocates for the increased use of IACI early in the disease course, whether as mono-therapy or in combination with other treatment modalities.

The PR-COIN registry during the study period encompassed 13 centers with a focus on care and outcomes improvement. Considering the generalizability of our findings, the demographic characteristics of our cohort are similar to demographics of other large North American databases, demonstrating that patient enrollment is likely representative of the JIA population (23). However, given the time period of the study, this dataset may under-represent medication use including IACI, as data collection of this type was not a primary focus of the registry at that time. The network emphasizes using data collected at the point of care, with data collection being performed on a voluntary basis. As such, data elements that directly capture outcomes such as the patient and provider assessment of disease activity are prioritized. While medication data such as IACI use is also captured, there are known gaps in the completeness of this data within the registry. Given that data entry and enrollment practices differ among centers, such variability could account for some of our findings.

It is also important to acknowledge that actual treatment decisions may differ from guidelines due to a number of factors,

including patient/family preferences and systems of care. For many pediatric patients, joint injections are performed under sedation so availability of sedation and space and time allocation for rheumatology procedures could be a factor. There may be center or regional differences in who is trained to perform injections. Ultimately, then, how closely actual treatments mirror what might be expected can be influenced by a number of factors beyond treatment guidelines. Such factors would ideally be captured in future work on this subject.

Since 2015, the PR-COIN network has continued to grow and has expanded efforts for data collection, incorporating standardized practices with electronic medical record (EMR) integration, which in the long term is likely to lend to a more clear understanding of medication use and potential associations to patient disease outcomes. This creates an opportunity for future research endeavors utilizing this robust registry. It also allows opportunity for collaboration. PR-COIN does not specifically intend to perform robust comparative effectiveness trials related to medications, though such trials are certainly needed to understand what treatment modalities might result in the best outcomes. PR-COIN is situated to help shed light on the gaps that need to be better studied and also to ultimately understand how implementation of recommended treatment strategies and quality improvement initiatives might affect outcomes over time.

4.1 Limitations

A notable shortcoming of this study is selection bias. Only patients with JIA who were enrolled in the PR-COIN registry were included, and it is possible that these patients are not representative of all patients with JIA. Additionally, there is very likely variability in registry enrollment patterns and data completeness between sites. At some centers, all providers help enroll patients while at other centers it may be only a limited number of providers who enroll their patients. Enrollment and data entry at the time of this study was primarily done through manual extraction, and there is no available information to capture what percent of the total JIA population from participating centers is represented by enrollment during this time frame. Furthermore, as previously noted, during this study period instances of IACI use for those in the registry may have been missed. This could be because IACI were given prior to registry enrollment, without retrospective treatment data necessarily being entered, and/or because only a short part of the patient course was captured in the registry, with over half of participants having been enrolled for less than a year. In any disease cohort, capturing the entirety of their course, including treatment given prior to enrollment and disease outcomes after treatment is an ideal but often difficult to achieve goal. Again, comprehensive, longitudinal treatment data and specific outcomes related to each treatment modality was not the primary objective for PR-COIN, though is of course desirable. This data set has additional limitations including lack of documentation of which joints were affected and/or injected and whether the same joint was injected repeatedly, which would lend to the depth and understanding of IACI use across the patient disease course. Further, limitations in available start dates for the various treatment modalities limited our ability to understand the relationships between treatments and their impact on disease control. Presumably, if a patient had well-controlled disease then joint injections and/or other treatment modifications would not be needed and so perceived underutilization of treatments could stem from our inability to determine disease activity and disease outcomes in relationship to treatment in this population.

In July 2015, PR-COIN transitioned to a new registry platform, creating some discontinuity in data. We therefore chose to analyze data only from the initial registry. Furthermore, in 2015 the production and availability of TH in the United States was discontinued; thus, comparing use of IACI before and after this time period could create limits in interpretation of IACI use and efficacy. We would predict a decline in IACI use in general after 2015. Another shortcoming, inherent to secondary analysis of existing data, is the fact that there may be additional confounding covariates that are not accounted for in the current data set.

4.2 Future directions

The PR-COIN network has continued to grow and expand since the period evaluated in this analysis and now encompasses over 8,500 patients with JIA. Transition to a new registry platform occurred in late 2022, with aspirations for more complete EMR integration. There are improvement efforts underway which will allow for more robust, detailed, and equitable data collection. As the network continues to focus on patient outcomes, standardization, and guideline-based care practices, this will allow for deeper examination of the relationship between treatment selection and duration of treatment in relation to clinical outcomes. In light of PR-COIN's focus on patient engagement, it will be important to consider how demonstration of treatment efficacy may influence treatment decisions. For example, patients and parents might wish to avoid systemic therapies when working to obtain disease control. Thus, a better and more direct understanding of the effectiveness of IACI compared to other treatments could influence a patient's acceptance of and adherence to recommended treatments. Further, a comparative analysis of historical and current IACI use practices may allow for deeper understanding of treatment practice changes over time and any potential relationship to patient disease outcomes.

5 Conclusion

In summary, this study highlights that the PR-COIN registry is a suitable representation of the North American JIA population, and analysis of registry data is able to provide meaningful insights on disease treatments utilized in patients with JIA. Within the registry from 2011 to 2015 we found that patients with persistent oligoarticular disease, ANA positivity, and use of other systemic medications were more likely to receive IACI. Utilization patterns also varied by treatment center. Overall, prevalence of IACI use was lower than might be expected, in particular for those with

oligoarticular disease, though the timing of our study in relation to published guidelines needs to be considered. Our findings highlight variability in treatment and that there are likely multiple factors contributing to treatment decisions throughout the disease course. Ultimately, understanding how these treatment decisions impact outcomes and whether standardization results in better outcomes is imperative. PR-COIN is well situated to shed light on this moving forward.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by the University of Minnesota Institutional Review Board (IRB) and not considered human subjects research. The studies were conducted in accordance with the local legislation and institutional requirements. The ethics committee/institutional review board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because this project was not considered human subjects research.

Author contributions

EB-D: Writing – original draft, Writing – review & editing. JW: Writing – original draft, Writing – review & editing. YG: Writing – original draft, Writing – review & editing. NR: Formal Analysis, Writing – original draft, Writing – review & editing. DB: Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2024.1423362/full#supplementary-material

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Assessing disparities through missing race and ethnicity data: results from a juvenile arthritis registry

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Introduction: Ensuring high-quality race and ethnicity data within the electronic health record (EHR) and across linked systems, such as patient registries, is necessary to achieving the goal of inclusion of racial and ethnic minorities in scientific research and detecting disparities associated with race and ethnicity. The project goal was to improve race and ethnicity data completion within the Pediatric Rheumatology Care Outcomes Improvement Network and assess impact of improved data completion on conclusions drawn from the registry.

Methods: This is a mixed-methods quality improvement study that consisted of five parts, as follows: (1) Identifying baseline missing race and ethnicity data, (2) Surveying current collection and entry, (3) Completing data through audit and feedback cycles, (4) Assessing the impact on outcome measures, and (5) Conducting participant interviews and thematic analysis.

Results: Across six participating centers, 29% of the patients were missing data on race and 31% were missing data on ethnicity. Of patients missing data, most patients were missing both race and ethnicity. Rates of missingness varied by data entry method (electronic vs. manual). Recovered data had a higher percentage of patients with Other race or Hispanic/Latino ethnicity compared with patients with non-missing race and ethnicity data at baseline. Black patients had a significantly higher odds ratio of having a clinical juvenile arthritis disease activity score (cJADAS10) of ≥5 at first follow-up compared with White patients. There was no significant change in odds ratio of

cJADAS10 ≥5 for race and ethnicity after data completion. Patients missing race and ethnicity were more likely to be missing cJADAS values, which may affect the ability to detect changes in odds ratio of cJADAS ≥5 after completion.
Conclusions: About one-third of the patients in a pediatric rheumatology registry were missing race and ethnicity data. After three audit and feedback cycles, centers decreased missing data by 94%, primarily via data recovery from the EHR. In this sample, completion of missing data did not change the findings related to differential outcomes by race. Recovered data were not uniformly distributed compared with those with non-missing race and ethnicity data at baseline, suggesting that differences in outcomes after completing race and ethnicity data may be seen with larger sample sizes.

KEYWORDS

health equity, data quality, juvenile idiopathic arthritis, learning health system, registry, electronic health record data

1 Introduction

Secondary use of electronic health record (EHR) data holds great potential for understanding patient populations, choosing interventions, and facilitating real-time research, overall pushing institutions toward becoming true learning health systems (1, 2). As we develop these learning health systems and large clinical and research databases, ensuring data quality becomes even more important (2). This is of particular importance in foundational areas on which further analyses will be performed, such as race and ethnicity data, especially given their known association with healthcare disparities.

While there is not a single standardized way of evaluating data quality, Feder has described a set of common domains that can be used to evaluate and improve data quality including data accuracy, completeness, consistency, credibility, and timeliness (2). The literature suggests three main threats to high-quality race and ethnicity data collection including accuracy, completeness, and consistency (3–5). Accuracy is defined as "the degree to which the value in the EHR is a true representation of the real-world value," completeness describes missing data, and consistency reflects truth of the value across multiple sources (2).

Reliable, culturally conscious ascertainment of race and ethnicity data, and completeness of entry are crucial for inclusion of minority populations in health systems' research and to mitigate inherent systemic bias (6–8). While race and ethnicity are social constructs, they serve as important markers for disparities and social determinants of health (9, 10). These concepts reflect a person's identity rather than a genetic or phenotypic basis, making self-reporting the gold standard for accurate race and ethnicity data.

Racial and ethnic minorities remain underrepresented in research despite similar willingness to participate (6). Incomplete race and ethnicity data can lead to exclusion from disparities analysis. Moreover, those missing this data are more likely to be Black or Hispanic, further worsening disparities and exclusion of minority patients from research (11, 12). Research and secondary analytics done with incomplete race and ethnicity can unintentionally worsen disparities (12–15). Alternatively, missing data may obscure disparities that are already present (12). Ensuring high-quality race and ethnicity data within the EHR and across linked systems, such as patient registries, allows identification of disparities and is necessary to achieve the goal of inclusion of racial and ethnic minorities in scientific research (3, 13).

We describe the iterative process of identifying and completing missing race and ethnicity data at six centers within the Pediatric Rheumatology Care Outcomes Improvement Network (PR-COIN). The PR-COIN database contains over 7,200 active patients with juvenile idiopathic arthritis (JIA) spanning 50,000 encounters with plans to add more pediatric rheumatologic diseases over time. Completing missing race and ethnicity data will help avoid unintentionally building inequitable algorithms and system structures. Furthermore, research done with incomplete data may make invalid inferences on disparities and stratification by race because of the exclusion of patients with missing data. This study provides a framework for addressing missing data and also explores the impact of filling in missing data on conclusions drawn from the registry.

2 Methods

This study was approved by the Seattle Children's Institutional Review Board and was conducted using data obtained through PR-COIN, collected by the physicians, providers, and families participating in this multicenter quality improvement collaborative (16).

This is a mixed-methods quality improvement study, consisting of the five following parts: (1) Identifying baseline missing race and ethnicity data, (2) Surveying current collection and entry, (3) Completing data (filling in missing race/ethnicity values) through audit and feedback cycles, (4) Assessing the impact of additional race and ethnicity values on outcome measures, and (5) Conducting participant interviews and thematic analysis. PR-COIN centers that were actively submitting data to the registry were eligible to participate. The eligible centers were issued an email invitation for voluntary participation in the research.

Baseline aggregate patient demographic and diagnosis data were obtained from the participating PR-COIN centers, and descriptive analyses were performed. The amount of missing race

and ethnicity data was calculated by center. Only patients present in baseline data were included in the subsequent rounds of data completion and final data analysis. We did not incorporate new patients enrolled into the registry during the study period. Due to the very small numbers of patients, three race categories independently defined in the registry were aggregated as "Other" for purpose of analysis, these were Asian, Native Hawaiian or Other Pacific Islander, and American Indian or Alaska Native. To maximize opportunities for data completion and accuracy, patients with designated registry categories of "Unknown," "Not Reported," and "Other" selected for race in the registry were aggregated with patients with the race field left blank to form the "Missing" category for requested completion. For ethnicity, any patients with registry categories of "Unknown" or "Not Reported" selected were aggregated with patients with the ethnicity field left blank to form the "Missing" category for this study. "Unknown" represents data not available in the EHR and "Not reported" represents patients who have chosen not to disclose their race and/or ethnicity.

A REDCap survey on race and ethnicity collection and upload methods was administered at each center prior to starting data completion and could be answered by the centers primary investigator, the research coordinator, or both. Survey questions are available in the Supplementary Material.

The survey included questions about race and ethnicity collection at the institution and methods of input into the EHR. Lastly, data were collected on race and ethnicity options within each EHR for comparison with registry options. The center with the lowest amount of missing data also notes use of race and ethnicity data in a "Master List." The Master List is a network recommended procedure in which centers create a list of all patients eligible for participation in the registry to monitor that registry enrollment is complete and reflective of the entire clinical patient population. Historically, the minimum data elements recommended for the Master List were patient name; medical records number (MRN); date of birth; gender; International League of Associations for Rheumatology (ILAR) code; diagnostic code; date of diagnosis; first, last, and next visit date; and provider; as described in a network Change Package (or instruction on keeping a Master List). Prior to this project, race/ethnicity was considered optional in construction of the Master List.

Audit and feedback cycles were performed by creating and sending reports of patients with "Missing" race and/or ethnicity data to each center. Centers were requested to complete the missing data fields within the registry using data already available in the EHR. After allowing a period for completion, new reports were generated and sent again with request for completion for a total of three cycles over 6 months. No new patients were added with the audit and feedback cycles, and any duplicate patient records were deleted from the registry. Data were obtained before completion (time 0), after round 1 of data completion (time 1), after round 2 of data completion (time 2), and after round 3 of data completion (time 3 or after completion). For round 1, centers were asked to focus on identifying and addressing any systematic reasons for missing data such as incomplete mapping or electronic transfer of data. If no such problems could be corrected, the center would manually complete data where possible. For round 2, centers were requested to manually fill in remaining missing data in the registry that was available in the EHR. For round 3, centers were requested to convert remaining "Missing" to either "Unknown" or "Not Reported," as appropriate. No patients were contacted for updating of race and ethnicity data.

We obtained clinical juvenile arthritis disease activity scores (cJADAS10) at first registry follow-up visit within 2–6 months of enrollment. cJADAS10 was chosen as an outcome measure owing to the prevalent use in the registry. It also contains components that are considered critical data elements with respect to data quality including patient global assessment, provider global assessment, and active joint count. Clinically, a low cJADAS10 indicated no or low disease activity and a high cJADAS10 indicated high disease activity with exact cutoff values varying by arthritis subtype (17). cJADAS10 is a continuous disease activity measure that is more sensitive to detecting change than the dichotomous American College of Rheumatology (ACR) criteria for inactive disease (17). We used a threshold of cJADAS10 $\geq$5 for all JIA subtypes using the cJADAS10 as this reflects greater than low disease activity for both oligoarticular and polyarticular arthritis. Odds ratio (OR) of cJADAS10 $\geq$5 at first visit after enrollment was compared before data completion and after data completion to assess how data completion changes the odds ratio of cJADAS $\geq$5.

We conducted two separate analyses: first using the initial data set with missing race/ethnicity values, and second with the updated data set that included observations with recovered missing values of race and ethnicity. For each analysis, we estimated the crude (univariable) OR of disease activity score, cJADAS10 $\geq$5, for age, gender, race, ethnicity, and JIA subtype. Then we used a multivariable logistic regression model to estimate the adjusted ORs for race and ethnicity, while accounting for differences between race and ethnicity groups in distribution of age and gender. Our interest was in the difference in ORs for race and ethnicity before and after recovering missing values of race and ethnicity. All analyses were performed in R studio.

Semi-structured, exploratory group interviews were conducted over two, 60 min virtual sessions with five out of six centers. The first interview had three participants from three centers and the second had five participants from four centers. Three centers had two participants in the interviews. The interviews were conducted to provide feedback on user experience with report format, to understand reasons for missing data, and identify best practice recommendations for completeness based on participant experiences. The participants had been involved in the data completion portion of the project and were known to the researcher prior to the interviews. The interview questions are available in the Supplementary Material. The first author and physician (KB) was the moderator and concurrently took notes during the interviews. The interviews were not recorded. They were followed by inductive thematic analysis conducted according to methodology and the steps outlined by Braun and Clarke and are described as follows (18). Coding was reviewed for agreement by a single second reviewer, another physician, and the last author on the paper, and any disagreement was resolved via discussion (EM).

1. **Familiarizing oneself with the data:** The notes from interviews were reviewed multiple times followed by a written summary and key points (KB).
2. **Generating initial codes**: The notes were reviewed line by line with codes assigned. Some lines were assigned multiple codes. This was performed twice with adjustment of codes during the second coding session (KB).
3. **Searching for themes**: The note segments were organized based on coding and used to identify themes or key concepts (KB).
4. **Reviewing themes**: The themes were compared with the interview questions and goals for alignment; both the reviewers established the themes (KB and EM).
5. **Define themes:** The meaning and patterns associated with themes and relationships between themes were identified. Discussion between reviewers was used to arrive at a consensus (KB and EM).
6. **Writing up:** The description of the themes is presented in the results section (KB).

3 Results

3.1 Identifying baseline missing data

A total of 2,359 patients with JIA were included across six PR-COIN centers. Table 1 depicts the demographics of the baseline population prior to data completion. At baseline, race was missing in 29% of the patients and ethnicity was missing in 31%. Of the 683 patients missing data on race, 669 (98%) of the patients were also missing data on ethnicity. The percentage of patients missing race or ethnicity data by center ranged from 0.5% to 99%. Patients with missing race data were more likely to be missing other metrics including ILAR subtype as well as cJADAS10 and its components. cJADAS10 was missing in 23% of all patients. Meanwhile, 50% of the patients with missing race or ethnicity data were also missing cJADAS, compared with around 12% of patients with non-missing race or ethnicity data at baseline. ILAR subtype was missing in 24% of all patients. Conversely, ILAR subtype was missing in over 50% of the patients with missing race or ethnicity data, while it was missing in only 12% of the patients with known race or ethnicity.

TABLE 1 Patient demographics.

Age	Frequency
Mean (SD)	11.4 (5)
Gender	
Female	1,653 (70%)
Male	706 (30%)
Race	
Black	105 (4%)
White	1,430 (61%)
Other	141 (6%)
Missing	683 (29%)
Ethnicity	
Hispanic/Latino	159 (7%)
Not Hispanic/Latino	732 (31%)
Missing	1,468 (62%)
ILAR code	
Oligoarticular (persistent and extended)	716 (30%)
Polyarticular (RF+ and RF−)	579 (25%)
Enthesitis-related arthritis	218 (9%)
Psoriatic arthritis	113 (5%)
Systemic JIA	109 (5%)
Undifferentiated arthritis	63 (3%)
Unknown	561 (24%)
Insurance	
Commercial/private	1,009 (43%)
Medicare/Medicaid	238 (10%)
Other	232 (10%)
Self-pay/none	163 (7%)
Missing	717 (30%)

SD, standard deviation; ILAR, International League of Associations for Rheumatology; RF, rheumatoid factor.

3.2 Survey of current collection and entry

Table 2 depicts the survey results. Registration was the primary staff for collecting race and ethnicity data for the EHR (5/6). Most centers (4/6) have a research coordinator that inputs data, including race and ethnicity data, into the registry. If race and ethnicity data are missing from the registry, no additional attempt is made to fill in that data in five of the six centers. One center cited difference in race and ethnicity categories between the institution and registry as a barrier to accurate data collection and entry. One center uploads data via electronic data transfer (EDT) from the EHR; all other centers enter the data manually. Data collection for the EHR occurs through a variety of methods across institutions including verbal reporting, direct entry online, and paper form. The center uploading data to the registry via EDT has the highest percent of missing race and ethnicity data compared with other sites because the demographic data were not mapped from the EHR to the registry fields. The center with the lowest amount of missing data also notes use of race and ethnicity in a "Master List."

All sites have the five minimum categories set by the National Institutes of Health (NIH) for race including American Indian or Alaska Native, Asian, Black or African American, Native Hawaiian or Other Pacific Islander, and White (9). The PR-COIN registration form includes these categories as well as Other, Unknown, and Not Reported with the ability to check multiple options to represent multiracial individuals. Two centers can select multiple races, four centers have Not Reported as an option, four have Other as an option, and Unknown is an option for one center. One center documents Hispanic/Latino as part of race, all others have a separate ethnicity category with Hispanic/Latino and Not Hispanic/Latino options.

3.3 Data completion via audit and feedback cycles

Throughout this section "baseline non-missing" will refer to patients whose race and ethnicity data were present before

TABLE 2 Center REDCap survey data.

Centers	A	B	C	D	E	F
Registry data entry method	Manual	Manual	Electronic data transfer	Manual	Manual	Manual
Registry data entry personnel	Not answered	Research coordinator, student	Research coordinator	Research coordinator, other	Other	Research coordinator
Master list?	Yes	No	Yes	Yes	Yes	Yes
Master list with race and ethnicity?	No	Not applicable	No	Yes	No	No
Master list updates	New enrollments	Not applicable	Monthly	Quarterly	Every other year	Weekly
Race/ethnicity data collection	Verbal collection	Direct entry, electronic form	Verbal collection, direct entry	Verbal collection, direct entry	Verbal collection, direct entry, paper form	Direct entry, paper form
Who inputs race and ethnicity in EHR?	Registration	Registration, other—parent	Registration	Unknown	Registration, scheduling	Registration
Who inputs race and ethnicity into PR-COIN?	Provider	Research coordinator, other	Research coordinator	Research coordinator	Other	Research coordinator
Is there a process for identifying missing race or ethnicity in PR-COIN?	No	No	No	No	No	Yes—demographic form at visit

completion. Percent baseline non-missing represents the proportion of a given race or ethnicity as a percent of the total patients without missing race or ethnicity data at baseline. Lastly, "recovered" represents patients with missing race or ethnicity data at baseline that were completed through audit and feedback.

Both missing race and ethnicity data decreased by 94% over the course of the project (from race missing in 29% of patients down to 2% missing and ethnicity missing in 31% down to 2%). Rounds 1 and 2 of the audit and feedback cycles showed the largest reductions in missing race and ethnicity data, as shown in Figure 1. There was a 45% decrease in missing race data after round 1. An additional 39% of missing race data were completed with round 2% and 10% in round 3. There was a 46% decrease in missing ethnicity data after round 1, a 33% decrease after round 2, and a 14% decrease after round 3. One center did not perform data completion during round 1 attributed to insufficient time to complete the task.

FIGURE 1
Percent change in race and ethnicity data by round of audit and feedback.

Figure 2 shows the distribution of race and ethnicity data as a percent of total patients, comparing before and after completion. The population distribution of race and ethnicity was consistent across all time points. The distribution of recovered race and ethnicity data is depicted by Figure 3. Recovered data were primarily White and Not Hispanic/Latino. "Deleted" represents patient entries that were identified as duplicate and deleted during the first round of data completion. Of those with race data that were recovered during the three rounds of audit and feedback, 63% were identified as White, 6% were identified as Black, and 11% were identified as Other (Figure 3A). Approximately 16% of patients were found to have duplicate entries, which were deleted. For patients with ethnicity data missing at baseline that was completed during the study, 64% were identified as Not Hispanic/Latino and 12% were identified as Hispanic/Latino (Figure 3B). Figure 4 shows the distribution of race and ethnicity data in patients as a percent of total patients with non-missing values at baseline and is compared with the race and ethnicity distribution in patients as a percent of total patients with recovered race and/or ethnicity. Race designated as Other was 55% higher in patients with missing race at baseline that was subsequently recovered (13%), compared with patients with non-missing race data at baseline (8.4%) (Figure 4A). Hispanic ethnicity was 50% higher in patients with missing ethnicity data at baseline that was subsequently recovered (15%), compared with patients with non-missing ethnicity data at baseline (10%) (Figure 4B).

Table 3 shows the change in missing data by center. Centers A–C and E had a completion rate of 98% or higher for race. Center F was able to complete two-thirds of their missing race. Center D decreased missing race data by 33%, decreasing patients missing race from three to two patients. Centers B–E completed data for 100% of those missing ethnicity. Center A decreased missing ethnicity data by 89% and center F decreased it by 66%. Of note, center C was missing 99% of race and ethnicity before completion and was also the only center uploading data to the registry via EDT.

3.4 Assessment of impact on outcome measures

3.4.1 Unknown cJADAS10

cJADAS10 from first registry follow-up 2–6 months after enrollment was obtained. Tables 4, 5 show the distribution of cJADAS10 ≥5, cJADAS10 ≤5, and unknown cJADAS10 before and after data completion for race and ethnicity, respectively. Before completion, 50% (341/683) of patients with missing race and 47% (341/732) with missing ethnicity had unknown cJADAS10. Meanwhile, cJADAS10 was unknown for 16% (17/105) of Black patients, 20% (28/141) of patients with Other race, and

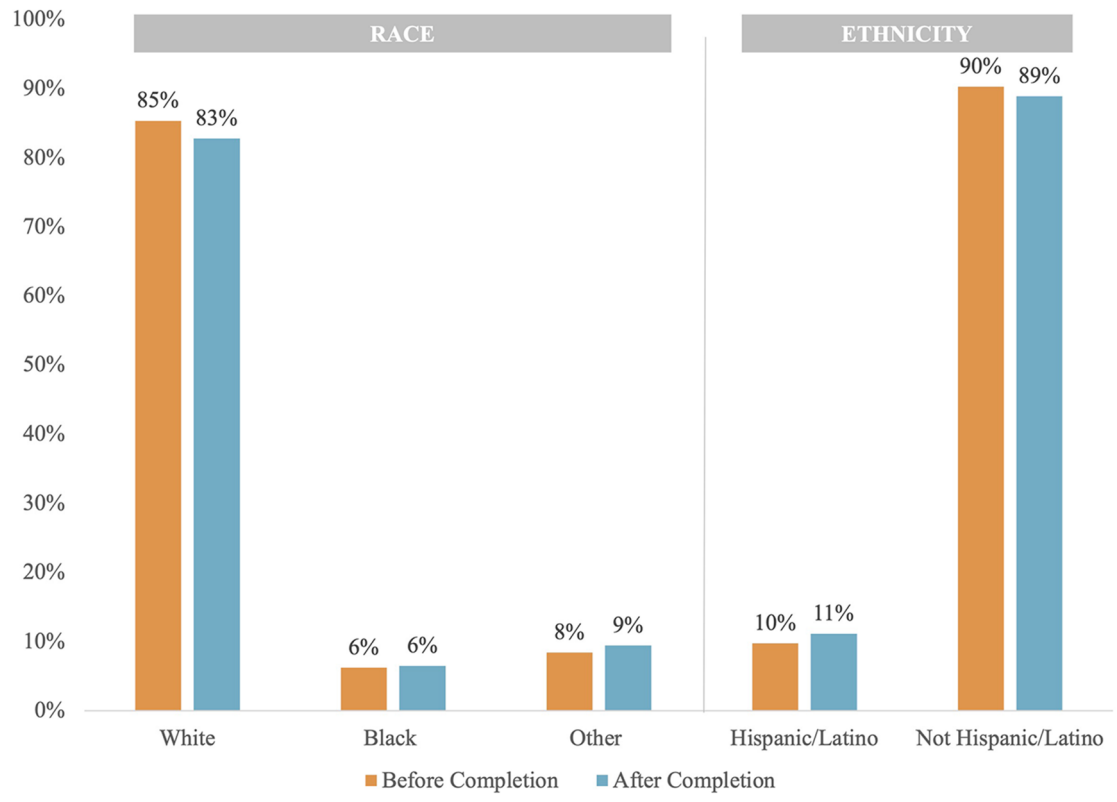

FIGURE 2
Population distribution of race and ethnicity data before and after data completion.

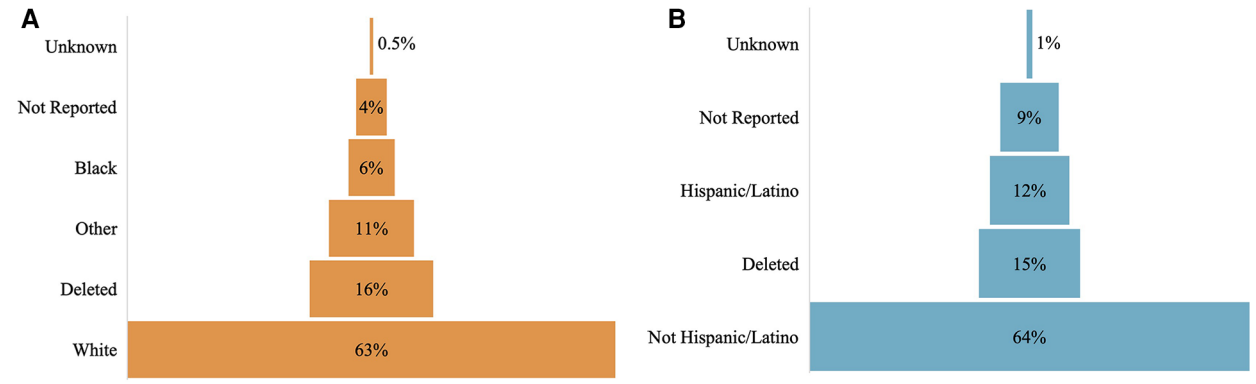

FIGURE 3
Distribution of missing data by race (A) and ethnicity (B).

12% (167/1,430) White patients. For ethnicity before completion, cJADAS10 was unknown in 16% (25/159) of Hispanic/Latino patients and 13% (187/1,468) of Not Hispanic/Latino patients.

Unknown cJADAS10 was seen more frequently in those with missing race data with 50% unknown cJADAS10 before completion and 49% unknown cJADAS10 after completion. Unknown cJADAS10 in those with missing ethnicity data increased from 47% to 65% from before completion to after completion. When race and ethnicity were known, unknown cJADAS10 ranged from 12% to 20% before completion and from 19% to 25% after completion.

3.4.2 Comparing cJADAS10 before and after completion

Tables 4, 5 also show cJADAS10 ≥ 5 for race and ethnicity before and after data completion. Before completion, cJADAS10 was ≥5 for 31% (438/1,430) of White patients, 41% (43/105) of Black patients, and 29% (41/141) of patients with Other race. cJADAS10 was ≥5 for 14% (97/683) of patients with missing race data and 15% (112/732) of patients with missing ethnicity data. For ethnicity data missing before completion, 30% (48/159) of Hispanic/Latino and 31% (459/1,468) of Not Hispanic/Latino patients had cJADAS10 ≥5.

After completion (round 3), cJADAS10 was ≥5 in 27% (494/1,834) of White patients, 28% (59/206) of Other patients, and 34% (49/144) of Black patients. CJADAS10 was ≥5 in 28% (67/239) Hispanic/Latino patients and 28% (528/1,910) Not Hispanic/Latino patients. The proportion of cJADAS10 ≥5 was decreased in all races and ethnicities after completion.

Patients with missing race data had the lowest frequency of cJADAS10 ≥5, present in 14% of patients before completion and 15% after completion. The findings were similar for those with

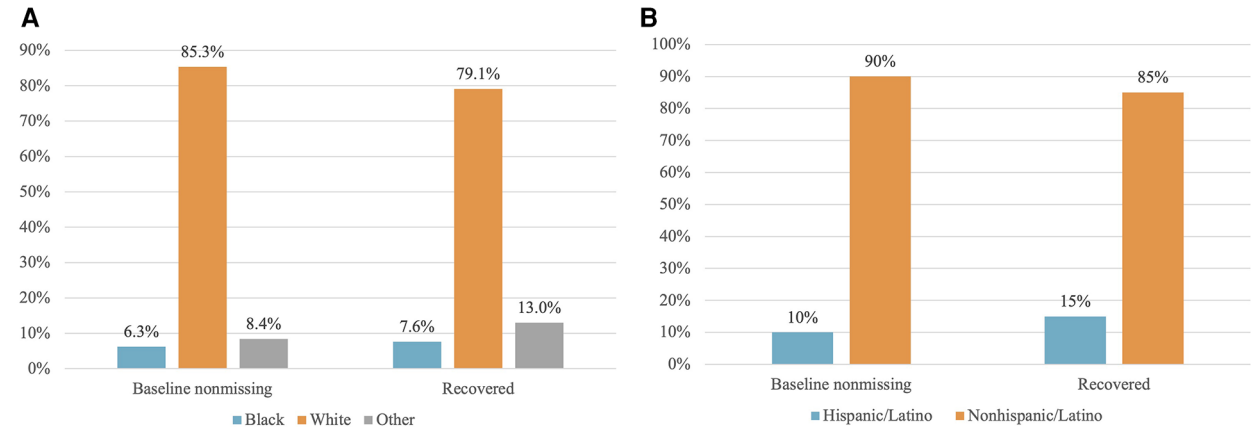

FIGURE 4
Distribution of patients with non-missing data at baseline vs. patients with data recovered. (A) Race: Baseline non-missing is the distribution of race data for patients whose race was present in the data set before completion. Recovered represents the distribution of race for patients whose race data were recovered and input into the registry during data completion, expressed as a percent of total patients with recovered ethnicity data. (B) Ethnicity: Baseline non-missing is the distribution of ethnicity in patients whose ethnicity was present in the data set before completion. Recovered is the distribution of ethnicity for patients whose ethnicity data were recovered and input into the registry during data completion, expressed as a percent of total patients with recovered ethnicity data.

TABLE 3 Missing data by center.

Centers	A	B	C	D	E	F
Missing race						
Before completion	47 (24%)	171 (37%)	248 (99%)	3 (0.5%)	160 (38%)	54 (13%)
After completion	1 (1%)	2 (1%)	18 (7%)	2 (0.3%)	0 (0%)	18 (4%)
Percent recovered	98%	99%	93%	33%	100%	67%
Missing ethnicity						
Before completion	70 (36%)	173 (38%)	248 (99%)	4 (0.6%)	166 (39%)	71 (18%)
After completion	8 (4%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	24 (6%)
Percent recovered	89%	100%	100%	100%	100%	66%

missing ethnicity data, cJADAS10 ≥5 was seen in 15% before completion and 14% of patients after completion. In patients with known race and ethnicity, 29%–41% had cJADAS10 ≥5 before completion and 27%–34% had cJADAS10 ≥5 after completion.

3.4.3 Odds of cJADAS10 ≥5

Table 6 presents the adjusted OR of cJADAS10 ≥5 at first registry follow-up for race and ethnicity comparing results before and after completion. The adjusted odds ratios control for patient age, gender, race, and ethnicity. Before data completion, the odds of cJADAS10 ≥5 were noted to be significantly higher for Black patients compared with White patients with odds ratio increased by 76% ($p = 0.011$). The odds ratio of cJADAS10 ≥5 for patients of Other races (OR = 1.12, $p = 0.596$) or those with missing race (OR = 0.97, $p = 0.916$) were not significantly different compared with White patients. The odds ratio of cJADAS10 ≥5 at first registry follow-up for Hispanic/Latino patients or those missing ethnicity were not statistically different from the odds ratios for Not Hispanic/Latino patients.

After data completion, controlling for patient age, gender, race, and ethnicity, the odds ratio of cJADAS10 ≥5 was significantly higher with a 61% ($p = 0.019$) increase for Black patients compared with White patients. The odds ratio of cJADAS10 ≥5 for patients of Other races (OR = 1.19, $p = 0.347$) or those missing race (OR = 1.39, $p = 0.352$) were not significantly different from the odds ratio of cJADAS10 ≥5 for White patients. For ethnicity after completion, the odds ratio of cJADAS10 ≥5 at first registry follow-up for Hispanic/Latino patients or patients missing ethnicity were not statistically different from the odds for Not Hispanic/Latino patients.

The estimated odds ratio for cJADAS10 ≥5 at first registry follow-up (2–6 months after enrollment) was higher for Black patients before completion compared with after completion. After completion the OR of cJADAS ≥5 decreased from 1.76 to 1.61, a relative decrease of 8.5%. The odds ratio of cJADAS10 ≥5 was not statistically significant when comparing White patients with patients with Other or missing race after data completion. The estimated OR of cJADAS10 ≥5 for Hispanic/Latino patients changed from 0.99 to 1.11, after data completion, a 12% relative increase. However, there was no statistically significant difference in the odds ratio of cJADAS10 ≥5 for Hispanic/Latino patients when compared with Not Hispanic/Latino patients.

TABLE 4 cJADAS10 distribution among race before and after completion.

	White	Black	Other	Missing race
Before completion				
cJADAS10 ≥ 5	438 (30%)	43 (41%)	41 (29%)	97 (14%)
cJADAS10 < 5	825 (58%)	45 (43%)	72 (51%)	245 (36%)
Unknown cJADAS10	167 (12%)	17 (16%)	28 (20%)	341 (50%)
After completion				
cJADAS10 ≥ 5	494 (27%)	49 (34%)	70 (30%)	22 (37%)
cJADAS10 < 5	999 (54%)	60 (42%)	112 (48%)	16 (26%)
Unknown cJADAS10	341 (19%)	35 (24%)	53 (22%)	22 (37%)

TABLE 5 cJADAS10 distribution among ethnicity before and after completion.

	Not Hispanic/ Latino	Hispanic/ Latino	Missing ethnicity
Before completion			
cJADAS10 ≥ 5	459 (31%)	48 (30%)	112 (15%)
cJADAS10 < 5	822 (56%)	86 (54%)	279 (38%)
Unknown cJADAS10	187 (13%)	25 (16%)	341 (47%)
After completion			
cJADAS10 ≥ 5	528 (28%)	67 (28%)	4 (14%)
cJADAS10 < 5	1,034 (54%)	115 (48%)	5 (21%)
Unknown cJADAS10	348 (18%)	5 (24%)	23 (65%)

3.5 Interviews analysis

Initial coding was performed by KMB based on interview notes. After the initial coding, both reviewers (KB and EM) established themes and resolved discrepancies via discussion to

TABLE 6 Odds ratio of cJADAS10 ≥5 for race and ethnicity before and after data completion.

Odds of cJADAS10[a] ≥5 before completion (N = 1,806)			Odds of cJADAS10[a] ≥5 after completion (N = 1,806)		
Predictors	Odds ratios	p	Predictors	Odds ratios	p
Ethnicity			Ethnicity		
Not Hispanic/ Latino	*Reference*		Not Hispanic/ Latino	*Reference*	
Hispanic/Latino	0.99	0.972	Hispanic/Latino	1.11	0.554
Missing	0.82	0.431	Missing	1.02	0.939
Race			Race		
White	*Reference*		White	*Reference*	
Black	1.76	**0.011**	Black	1.61	**0.019**
Other	1.12	0.596	Other	1.19	0.347
Missing	0.97	0.916	Missing	1.39	0.352

[a]cJADAS10 is defined as cJADAS10 score ≥5 at the first registry follow-up visit (2–6 months after enrollment).
Bold values indicate statistical significance ($p < 0.05$).

establish the final emergent themes. Three themes emerged from the inductive thematic analysis of the post-completion interview sessions including project experience, variation in reporting and data collection, and defining data processes. We also gathered participant recommendations with regards to improving data collection moving forward.

3.5.1 Project experience

For project experience, the participants noted that the data completion process was manageable and sustainable. Use of an audit report was noted to be helpful in identifying and completing missing race and ethnicity data. Most sites completed registry data via the demographics data present within the EHR entered during the clinic registration process. Three centers reported that portions of missing data were not able to be identified within the EHR. Duplicate data were identified in one site resulting in working with the registry platform for resolution. Another center worked with the registry platform manager, to troubleshot EDT and data migration issues. One center initiated a site-specific quality improvement project to educate staff on appropriate collection and self-reporting of race and ethnicity data.

3.5.2 Variation in reporting and data collection

Multiple centers noted confusion and inconsistent documentation practices around "Unknown" vs. "Not Reported" as options and appreciated education around this distinction, recommending adjustment of these terms within the registry. One center noted that many marked as "Not Reported" had data present within the EHR. Meanwhile, another center hypothesized that their large number of "Unknowns" may reflect a lack of options with which a patient identified. The separation of Hispanic/Latino ethnicity from racial groups is also noted as an area of confusion for some patients. One center also documents Hispanic/Latino as race, which can result in difficulty with data reconciliation as the patient may not identify a race category separate from their ethnicity. Multiracial is also a source of difficulty for data mapping, multiple centers have multiracial as a single select option. PR-COIN allows for multiselect to document two or more races but does not have a multiracial, single select option. The centers also noted ongoing changes in their data collection practices including processes and options that result in ongoing challenges for data mapping and upload.

3.5.3 Defining data processes

Many centers commented on the lack of understanding or transparency of the institutional race and ethnicity data collection practices. Multiple centers used this project as a starting point for improving overall registry data entry, staff education, as well as understanding and improving data collection practices at the institution level. The center uploading via electronic data transfer identified that race and ethnicity were not part of the transfer, resulting in 99% missing race and ethnicity. Strategies for manual verification were suggested including using a site Master List with race and ethnicity to identify those missing data and frequent audits of race and ethnicity for new enrollments.

3.5.4 Participant recommendations

1. Race and ethnicity should be considered critical data elements.
2. Adjustment of wording for Unknown and Not Reported options to improve consistency with documentation.
3. Develop a tip sheet on best practices for race and ethnicity data collection and entry.
4. Identify which elements are/are not included in electronic data transfer.

4 Discussion

Among the six participating centers, a mean of one-third of race and ethnicity data was missing within the PR-COIN registry, with substantial variability across centers. This mean number is consistent with previous reports of missing race and ethnicity data in other databases (12, 13, 19). When considering use of patient registry data for disparities research or equity-related quality improvement, complete and accurate data are important to prevent exclusion of these patients in analysis due to missing data. This project has demonstrated that race and ethnicity data quality can be improved through manual completion from the EHR where most of the missing data can be found. In this scenario, data can be improved via audit and feedback cycles through EHR data, which may ultimately lead to improved completion of the race and ethnicity data. Future, registry-wide data completion efforts could reasonably be completed in one to two rounds given signs of diminishing returns for this cohort after the second round of completion.

We recommend that race and ethnicity data be critical data elements with the PR-COIN and all registry frameworks. This could eliminate a large amount of missing data at the registry level without significant additional work from a data collection standpoint. For example, this may mean that registration cannot be completed without race and ethnicity data, prompting sites to perform the extra step of looking up this information in the EHR. In addition, we recommend ongoing data auditing and improvements. This could be accomplished via the Master List by adding race and ethnicity data to create a self-reporting mechanism to maintain data completion.

Previous reports have suggested that missing data are often disproportionately Black and Hispanic/Latino (11, 12). We found higher proportions of Hispanic/Latino ethnicity and Other races in recovered data compared with the baseline population of patients with non-missing race or ethnicity. However, the population distribution remained stable. Given the slightly skewed distribution of recovered data, additional data completion at a larger scale may reveal changes in the population distribution. However, given the concordance between missing race and ethnicity and other missing data elements such as cJADAS10 and its components, missing race and ethnicity data may identify patients with larger data quality problems.

While other studies have identified new or worsened disparities with completion of race and ethnicity data, we found no difference in the odds ratio of having a cJADAS10 $\geq$5 at first registry follow-up after data completion. This may be due to the near uniform

distribution of patients with missing race and ethnicity data. However, 50% of the patients with missing data were also missing cJADAS10. It is possible that, due to this missing data, we could still be missing small changes in disparities assessments for cJADAS10. Although there was not an identified impact on our outcome assessment before and after data completion, the completion of this data remains an important priority. As a result of this effort, there are now over 600 patients with completed race and/or ethnicity data that will be included in future disparities assessments.

This project has informed improvements and best practice recommendations for the registry moving forward. Multiple centers have embarked on formal or informal education and quality improvement initiatives to understand and optimize data collection into the EHR and entry into the registry. These are the first steps to determine data accuracy that must be validated and improved at each institution. We identified that the center entering registry data via EDT was missing 98% of race and ethnicity due to data mapping and transfer issues. Mapping issues also exist for centers with manual entry due to discordance between registry options and options for race and ethnicity. Specifically, Hispanic/Latino and multiple races, via multiselect or single select options, are noted to increase difficulties with data reconciliation, which can compromise data accuracy. There is ongoing work for standardization and implementation of race and ethnicity data collection along with other social determinants of health, which may provide helpful guidance for data mapping in the future (20). Moving forward, we can recommend that race and ethnicity be included as critical data elements to prioritize input during registration and provide ongoing data quality feedback.

As of March 2024, the Office of Management and Budget (OMB) standards has published new recommendations for race and ethnicity data with two major changes: (1) Hispanic/Latino will now be part of race with no ethnicity category. (2) There will be an additional minimum racial category of Middle Eastern or North African, which may similarly provide mapping and data challenges across different centers as these new recommendations are implemented across different institutions (21). This has implications that registries may need to consider on future data capture, especially if health systems update their collection of this data into the EHR to reflect these changes. These updates also serve as a reminder that race and ethnicity are social constructs and the categories offered are an incomplete representation of these concepts. Completeness is just the first step in having robust data in this space. Accuracy and reliability are also incredibly important but hard to achieve amidst an incomplete and changing framework for race and ethnicity data. Thus, we also recommend having a system in place to continually review and update how the data are collected and what options are offered. Opportunities for patients to self-identify are important to ensure we are representing our patients as accurately as possible.

When using a registry or learning health system to monitor and address disparities, having complete race and ethnicity data is extremely important for accurate assessments. Prior to data completion, disparities assessments would have excluded almost one-third of patients due to missing data. Thus, learning health systems with missing race and ethnicity data are at risk of widening disparities through exclusion from research and inaccurate assessment of disparities. Addressing race and ethnicity data quality should be a component of equity work within learning health systems. This project provides a baseline assessment of missing data and outlines a data completion process that can be applied to all centers and new disease additions to the registry moving forward.

Data availability statement

The data analyzed in this study are subject to the following licenses/restrictions: Data use and legal agreements present for use of PR-COIN data restrict public availability of data. Requests to access these data sets should be directed to Jade Singleton, jade.singleton@seattlechildrens.org.

Ethics statement

The studies involving humans were approved by the Seattle Children's Hospital Institutional Review Board. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

KB: Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Project administration, Visualization, Writing – original draft, Writing – review & editing. JS: Data curation, Formal analysis, Methodology, Writing – review & editing. XW: Formal analysis, Supervision, Writing – review & editing. SV: Writing – review & editing. JH: Writing – review & editing. AL: Writing – review & editing. NP: Writing – review & editing. JK: Writing – review & editing. DF: Writing – review & editing. EH: Writing – review & editing. MG: Writing – review & editing. CK: Writing – review & editing. AM: Writing – review & editing. PT-H: Supervision, Writing – review & editing. EM: Conceptualization, Funding acquisition, Investigation, Methodology, Project administration, Supervision, Validation, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this paper can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2024.1430981/full#supplementary-material

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Using a collaborative learning health system approach to improve disease activity outcomes in children with juvenile idiopathic arthritis in the Pediatric Rheumatology Care and Outcomes Improvement Network

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Abbreviations

ACR, American College of Rheumatology; CARRA, Childhood Arthritis and Rheumatology Research Alliance; CCM, chronic care model; cJADAS10, 10-joint clinical juvenile arthritis disease activity score; EHR, electronic health record; IRB, Institutional Review Board; JIA, juvenile idiopathic arthritis; LHN, learning health network; PWG, parent working group; PAT, patient advocacy team; PM, population health management; PR-COIN, Pediatric Rheumatology Care and Outcomes Improvement Network; QI, quality improvement; QM, quality measure; SMS, self-management support.

Introduction: The Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) is a North American learning health network focused on improving outcomes of children with juvenile idiopathic arthritis (JIA). JIA is a chronic autoimmune disease that can lead to morbidity related to persistent joint and ocular inflammation. PR-COIN has a shared patient registry that tracks twenty quality measures including ten outcome measures of which six are related to disease activity. The network's global aim, set in 2021, was to increase the percent of patients with oligoarticular or polyarticular JIA that had an inactive or low disease activity state from 76% to 80% by the end of 2023.
Methods: Twenty-three hospitals participate in PR-COIN, with over 7,200 active patients with JIA. The disease activity outcome measures include active joint count, physician global assessment of disease activity, and measures related to validated composite disease activity scoring systems including inactive or low disease activity by the 10-joint clinical Juvenile Arthritis Disease Activity Score (cJADAS10), inactive or low disease activity by cJADAS10 at 6 months post-diagnosis, mean cJADAS10 score, and the American College of Rheumatology (ACR) provisional criteria for clinical inactive disease. Data is collated to measure network performance, which is displayed on run and control charts. Network-wide interventions have included pre-visit planning, shared decision making, self-management support, population health management, and utilizing a Treat to Target approach to care.
Results: Five outcome measures related to disease activity have demonstrated significant improvement over time. The percent of patients with inactive or low disease activity by cJADAS10 surpassed our goal with current network performance at 81%. Clinical inactive disease by ACR provisional criteria improved from 46% to 60%. The mean cJADAS10 score decreased from 4.3 to 2.6, and the mean active joint count declined from 1.5 to 0.7. Mean physician global assessment of disease activity significantly improved from 1 to 0.6.
Conclusions: PR-COIN has shown significant improvement in disease activity metrics for patients with JIA. The network will continue to work on both site-specific and collaborative efforts to improve outcomes for children with JIA with attention to health equity, severity adjustment, and data quality.

KEYWORDS

juvenile arthritis, quality improvement, outcome measures, pediatrics, rheumatology, registries, collaborative learning

Introduction

Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) is a learning health network (LHN) designed to improve and advance the care of children with juvenile idiopathic arthritis (JIA) (1, 2). JIA is a chronic autoimmune disease affecting about 1 in 1,000 children that can lead to life-long damage to joints from arthritis and vision loss from uveitis without proper care. LHNs leverage multisite stakeholders including patients, families, medical providers, other healthcare staff, researchers, and community organizations working together with a common goal and sense of urgency to develop knowledge from data to deliver better clinical care and improve health outcomes more equitably.

Using methodology from the Model for Improvement, the Institute for Healthcare Improvement Breakthrough Series, and with quality improvement (QI) guidance and initial coordination from the James Anderson Center for Health Systems Excellence at Cincinnati Children's Hospital Medical Center, PR-COIN modeled its beginnings after ImproveCareNow, a pediatric inflammatory bowel disease multi-center collaborative of currently over 100 participating medical centers for its significant achievements including sustained remission rates in their patient population (3–5). Eager to achieve similar improvements in JIA outcomes, PR-COIN launched in 2011 as an improvement collaborative with an inaugural membership of 12 centers and started its journey to achieve extraordinary rates of disease control in JIA while using clinical data for QI and research with a goal to accurately and reliably measure and report performance on process and outcome quality measures to drive improved outcomes (1).

JIA is a lifelong disease with a high risk of morbidity related to both the disease and its treatments, potentially causing permanent damage to joints and eyes. Early diagnosis and timely, effective treatment are crucial as JIA can significantly impact a child's growth, development, and quality of life (6). A 17-year follow-up study of patients with JIA revealed a generally favorable outcome for most patients, yet ocular involvement remained prevalent (7).

Despite good physical and social functioning, many patients expressed feeling burdened by their condition, with current disease activity strongly influencing functional status. Predictors of long-term active disease include early onset, specific joint involvement, and elevated inflammatory markers (8).

Over the past two decades, several outcome measures have been developed and validated to monitor how JIA progresses and to help manage it effectively at the point of care. These measures are designed to provide a comprehensive view of a patient's condition, that allow for tailoring treatments to individual needs and monitoring overall disease progression and response to therapy. Key measures focus on clinical disease activity, functional status, radiographic outcomes, laboratory markers, and patient-reported outcomes.

Utilization of outcome measures is essential because it enables providers to better track disease progression, assess treatment efficacy, effectively monitor disease progression, and implement timely interventions for better outcomes. PR-COIN utilizes QI methodologies to enhance collection and monitoring of outcome measures in JIA. By systematically analyzing and improving the care processes, PR-COIN aims to enhance the effectiveness and efficiency of JIA management.

PR-COIN employs various QI strategies, such as Plan-Do-Study-Act cycles, to iteratively test and refine changes in clinical practice. Through collaborative efforts among healthcare providers, researchers, and patients, PR-COIN identifies areas for improvement in the utilization of outcome measures in clinical care, such as enhancing the sensitivity of detection, standardizing assessment methods, and integrating patient-reported outcomes. By incorporating feedback from stakeholders and continuously evaluating the impact of interventions, PR-COIN ensures that improvements in outcome measures are evidence-based and patient-centered.

Moreover, PR-COIN leverages data-driven approaches to monitor progress and benchmark performance across different healthcare settings. By collecting and analyzing real-world data on JIA outcomes, PR-COIN identifies best practices and facilitates knowledge sharing among participating institutions. This collaborative learning environment accelerates the dissemination of effective strategies for enhancing outcome measures in JIA care.

PR-COIN has a shared patient registry that currently tracks 20 quality measures (1). Quality measure categories include outcome, process, balancing, and data quality measures. PR-COIN has ten quality measures measuring health care outcomes including six related to disease activity and four patient-reported outcomes. The focus of this manuscript is reporting of the disease activity outcome measures. The PR-COIN collaborative's global aim in 2021 was to increase the percent of patients with oligoarticular or polyarticular JIA in an inactive or low disease activity state from 76% to 80% by the end of 2023.

Materials and methods

This manuscript utilized the SQUIRE 2.0 reporting guidelines (9).

Context

PR-COIN uses a collaborative learning health system approach to improve quality of care and outcomes for children with JIA (10–12). PR-COIN currently has 23 participating sites from academic pediatric medical centers throughout the United States and Canada. PR-COIN is led by a coordinating center which provides quality improvement consultation, quality improvement education, maintenance of certification opportunities, data management, data analytics, legal and regulatory supervision, project development and oversight, and overall support to the network. Additionally, PR-COIN has seven operating committees directing Measures, Outcomes, Informatics, Scientific Development and Oversight (Research), Engagement, Finance and External Partnerships, and Education activities all led by volunteer members. The leaders for each committee together form the Executive Committee along with the principal investigator to prioritize network-wide initiatives in line with the stated mission and vision of PR-COIN (2). Elected members join the committee leads to form the Steering Committee to provide additional representative network oversight.

PR-COIN member centers have local QI teams that vary in composition by site, but typically consist of a physician champion, other providers including rheumatologists, pediatric learners (e.g., rheumatology fellow, pediatric resident, medical student), occupational and physical therapists, nurses, and other staff including medical assistants, social workers, administrative staff, and research staff. Some centers receive local QI improvement specialist support from their institution. Most valuable is the personal contribution of patients and families to PR-COIN QI work at both the local team and network committee level lending their experience and expertise. Patients and families contribute to workgroups of specific interests, educational presentations, development of QI tools and other items dealing with specific challenges unique to the JIA population. Local team members conduct QI projects of greatest value to their site using the Model for Improvement and rapid plan-do-study-act cycles, contribute to network led initiatives, and "share seamlessly and steal shamelessly" the best practices presented at monthly action-period calls and twice-yearly learning sessions held in person and virtually to accommodate participation from all members.

Data from PR-COIN sites are collected at the point-of-care, with the goal to collect data on every patient at every visit, in order to calculate performance on JIA quality measures. PR-COIN has a shared registry platform, operated by vendor Hive Networks, allowing individual sites to see both site and aggregate data in a centralized platform to monitor quality measure performance (1, 13). The PR-COIN registry contains data from over 13,500 registered patients, over 7,200 of whom are active patients, and greater than 89,000 patient visits.

Interventions

QI tools

The collaborative utilized QI tools in their improvement efforts including creation of a key driver diagram (Figure 1) to identify

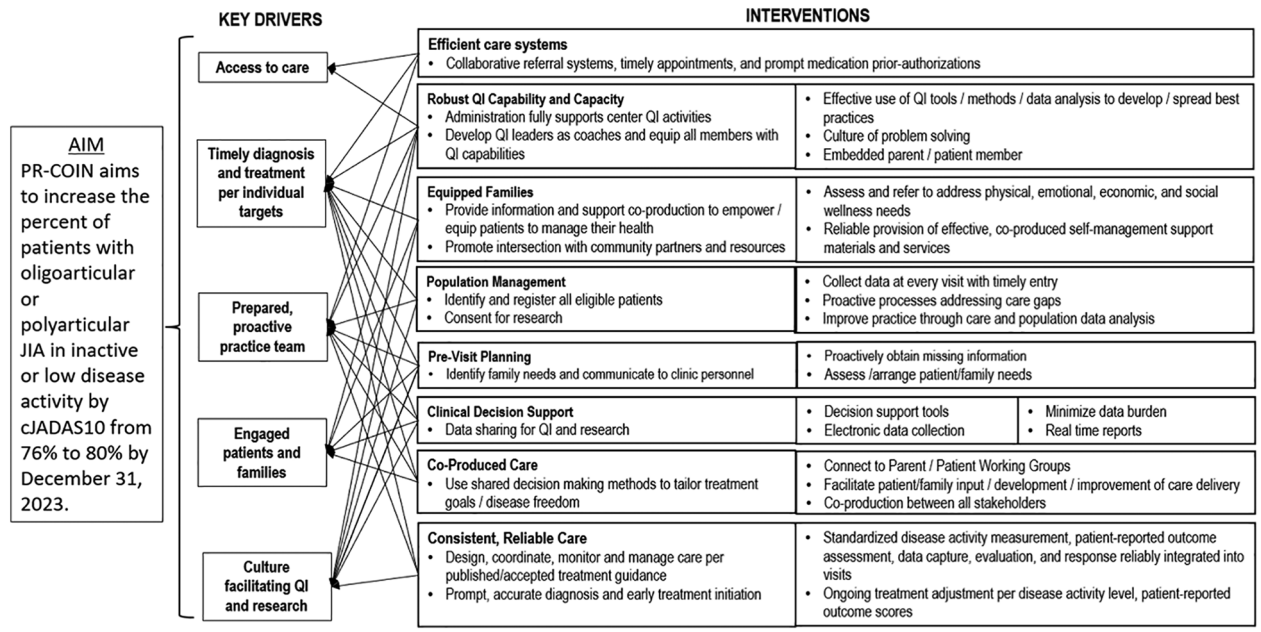

FIGURE 1
Key driver diagram highlighting our aim, primary drivers, and potential interventions.

drivers and interventions to help achieve their aims. The collaborative and many sites also utilized other QI tools including process maps, cause and effect diagrams, failure modes and effects analyses, and pareto charts. PR-COIN sites have conducted numerous interventions to help improve performance on quality measures, including disease activity outcome measures. Interventions have been both site-specific and network-wide. Some interventions or interventional themes have spanned multiple sites as PR-COIN has facilitated several network-wide initiatives.

PR-COIN uses strategies advocated by the Chronic Care Model (CCM) (14). The CCM is an organizational approach to delivery of healthcare for chronic diseases and includes six key domains in which high quality health care can be developed through QI efforts including the community, the health system, self-management support, delivery system design, clinical decision support, and clinical information systems (14). Studies suggest employment of the CCM improves healthcare delivery and outcomes for patients with chronic diseases (15, 16).

Pre-visit planning

One early network intervention adopted by PR-COIN was the use of pre-visit planning (PVP) (17). PVP is the process whereby the clinical team reviews the electronic health record (EHR) and may also survey patients to make sure that the data that are needed for the clinic visit is readily available at point of care (18). When health care teams are prepared for clinic visits, valuable patient-facing time in clinic is not wasted on tracking down results or reviewing prior medical records. Care gaps can be pre-identified and addressed at that visit. In the setting of juvenile idiopathic arthritis care, this includes having recent lab data and ophthalmology uveitis screening reports available as well as most recent arthritis disease activity scores. Automated PVP reports can be generated from existing data in the PR-COIN registry so not every item has to be manually collected. As pediatric rheumatology sites onboard to PR-COIN, they are encouraged to implement PVP such that it becomes standard in their practices. Effective implementation of PVP saves time for each patient, thereby increasing practice efficiency.

Population health management

Population health management (PM) is an approach that aligns with the PR-COIN mission to achieve equitable care and close gaps in care to improve quality measure (QM) performance (19–22). The intent is to leverage clinical information systems (electronic data transfer from EHR systems into the shared registry) to be able to generate reports looking across the entirety of patients in the registry (population), including reports of individual patients who "fail" to pass a measure to prompt action. PM is also critical to avoid loss to follow-up care, particularly of the most vulnerable patients with ongoing active disease. For example, if the goal is to achieve low disease activity or inactive disease, the registry reporting feature can be used to drill down to identify patients with moderate or higher disease activity. A local care coordinator can then conduct outreach based on the reports, e.g., contact patients to schedule visits in case of loss to follow-up and high risk (e.g., moderate disease activity and not seen for >180 days) to be sure treatment is adjusted if the condition is still not under adequate control. PM is an efficient and reliable way to ensure care standards are met across a population. To establish effective PVP, PM at the local hospital level may result in delivery system design changes as part of successful implementation.

Shared decision making

A key tenet of the CCM is that disease outcomes will be superior if the patients are invested and engaged in their own healthcare. This led to shared decision making as another network-wide intervention espoused by our LHN (23, 24). PR-COIN developed medication issue cards as a tool for shared decision making to assist patients and families in having discussions with providers that inform selecting their preferred medication regimen to treat their arthritis (25–29). The decision cards focus the discussion on aspects of a medication most important to patients/families such as side effects, frequency of administration, cost, and other factors. In addition to increasing patient engagement, this approach ensures the health care delivery is patient-centered, which is another key element of the CCM. PR-COIN sites have access to these medication issue cards and can utilize them in discussing arthritis medication initiation or changes in therapy.

Self-management support

Self-management support (SMS) is the act of empowering or facilitating patients and their family's ability to successfully manage their own medical condition on a day-to-day basis (30, 31). This would incorporate regular assessment of barriers to care and treatment, assistance with finding solutions to problems, and the setting of patient goals with follow up on progress in achieving those goals. PR-COIN launched a network-wide SMS initiative where site members were trained on SMS tools including motivational interviewing, and PR-COIN sites were encouraged to conduct QI work around introducing SMS into practice (32). PR-COIN developed several SMS tools to assist pediatric rheumatology providers, including a SMS change package (33). PR-COIN also adapted The Helping Hands Handbook from Cincinnati Children's Hospital Medical Center. This handbook was created by patients and families with JIA and pediatric rheumatology providers to assist patients and families on their journey navigating life with JIA. This handbook provides information on a wide array of JIA-related topics in limited-literacy and patient-friendly language including education about different aspects of the disease, medications, school accommodations, vaccine considerations and many other components. In addition, PR-COIN team members in conjunction with other researchers created a SMS tool called the barriers assessment tool, which asks the patient/parent to check off different barriers to taking medications including side effects, cost of medication, worry about side effects, forgetting to take medication, and more (33). This tool asks patients to consider these barriers for oral, subcutaneous, and infusion medications as well as barriers to completing occupational and physical therapy. Patients and providers have found the barriers assessment tool helpful at uncovering barriers to care that otherwise might have gone unaddressed. This tool drills down to the root cause of nonadherence to taking medication, which is a problematic aspect in managing chronic diseases such as JIA.

Parent and patient engagement

Patient and parent engagement in PR-COIN reflects a commitment to inclusion of all LHN stakeholders in governance, participatory leadership, and in creating a structure for healthcare improvement with quality measures (QMs) that are accountable to patients (34). Parent involvement in co-creation and governance of the network as partners with clinicians and researchers has resulted in a network that has at its core a focus on patient outcomes, and in its heart a focus on meeting the needs of patients and their families. In the goal to be patient-centered, the network has embraced a co-production approach to ensure that the product (delivery of exceptional and equitable health care service and meaningful research) is responsive to the priorities and needs of the patients and families (35). Parents lead and comprise the PR-COIN Engagement Committee, with its associated Parent Working Group (PWG; a parent advisory council) and Patient Advocacy Team (PAT). The PWG/PAT inform and develop patient and family facing educational materials for patient learning and empowerment to foster self-management, reduce barriers to care, and generate tools to enable shared decision making. The parents create public awareness materials (social media, videos) to communicate the work of the network to garner community support and participation.

Parents play a vital role in fostering empathy within the network. Communication of the patient experience is critical for clinicians to become knowledgeable to the impact of health care activities, disease, and its treatment on a personal level. This communication occurs in a manner that is absent or incomplete in the clinic exam room, in which a differential power dynamic, lack of time or other factors may prevent the full disclosure of the scope of disease impact to the clinician. In PR-COIN, there is deliberate intent to remove the hierarchical structure of physician-patient interactions and cultivate a collaborative decision-making setting. Parents and patients are invited to participate as equal partners in all PR-COIN committees bringing the patient perspective to inform and shape network operations and activities and to help set research priorities. Parents present "ignite talks", create and administer surveys of patient and families to garner broad representative input on topics of network interest, and share and instill the patient voice in network learning sessions and conferences.

The ImproveCareNow LHN has proposed 5 metrics of engagement of patient advisory councils, namely: (1) that there be personal growth for members, (2) internal engagement in community, (3) presence within the LHN, (4) engagement at the local center level, and (5) members contribute to products (36). All of these areas are encouraged in PR-COIN, although engagement at the local center level occurs with variable success. As children move through the system, and invariably transition to adult care so is the need to periodically recruit new parents to work with teams. It can be challenging to meaningfully involve parents into local improvement work, due to their own/family competing interests and job duties during traditional working hours when health care teams meet.

Batalden et al. describe the concept of healthcare as a co-produced service with patients. Likewise, for the LHN model to be effective in design, it requires that it be co-produced by stakeholders, of whom the parents and patients are central (35). In order for PR-COIN to achieve the stated mission that was formulated with parent input, the parents and patients will continue to be involved and represented in the design and measurement of LHN interventions informed in part by their lived experiences. The interventions comprised in the CCM, especially self-management support, underscore the idea of health as a co-produced service. PR-COIN work in the area of shared decision making reflects the steadfast approach of the network towards parents engaged as true partners in care. The PR-COIN registry platform enables parent committee members equal access to shared materials and collaborative files, with protected health information and patient data under separate protection.

While the fundamental drive of LHNs is to reduce unwarranted variation in care to reduce care gaps, increase safety, and promote health equity, the tension with shared decision making and co-production of care is that variation in care may re-enter at the patient level intentionally and according to patient preference (35). This drives home the importance of accurate, health literate and numerate materials to support patient and families to be empowered in informed decision making.

Treat to target

"Treat to Target" is an intervention approach that serves to anchor co-production to shared goals of care of the clinician and family (37–39). In this setting, parents select a target for care, classically, "inactive disease" or "low disease activity". The clinician then works with the family according to guidelines for a Treat to Target approach, which involves systematic assessment of a disease activity measure at regular intervals to allow for adjustment of medication towards reaching the parent/patient goals on disease control or other individual goal (37). A consensus meeting with clinicians and parents highlighted the importance of the patient being able to establish their individual treatment goal, and that it be tracked over time as the treatment plan was adjusted to meet this and other identified goals of care, e.g., disease control, pain control, physical activity, school attendance, etc (38).

Study of the interventions

PR-COIN regularly reviews QM performance. Select measures are often highlighted during monthly "action period calls". Furthermore, a deeper dive into the data is done twice a year during the network's "Learning Sessions" when we review measure performance and highlight best practices among sites. Attendees include providers, nurses, other clinical staff, research coordinators, patients/parents, informatic specialists, and registry staff. Additional data review is done at various intervals at a site level, at the coordinating center, during maintenance of certification cycles, and at different committee meetings.

Measures

PR-COIN has a complete QM set with disease activity outcomes, patient-reported outcomes, process measures, data quality measures, and a balancing measure (1). There are six outcome measures related to disease activity. Our primary outcome measure is patients with oligoarthritis or polyarthritis who have inactive or low disease activity by the 10-joint clinical Juvenile Arthritis Disease Activity Score (cJADAS10). The oligoarthritis group includes patients with the persistent oligoarticular subtype. Polyarthritis includes patients with the International League of Associations for Rheumatology subtypes of extended oligoarticular and polyarticular (both rheumatoid factor negative and positive). The cJADAS10 cut-offs to define inactive or low disease activity for each group were established from the literature, and the cJADAS10 value from the patient's last visit is used (40). Inclusion criteria includes a patient having at least two clinic visits with a clinic visit in the past 450 days. Patients are excluded if they are missing one or more component of the cJADAS10—physician global assessment of disease activity, patient/parent global assessment of overall wellbeing, and active joint count. Another similar measure is the inactive or low disease activity by cJADAS10 by 6 months (after diagnosis). This measure uses the same JIA subtypes and disease activity cut-offs. However, the denominator only focuses on patients recently diagnosed (180–270 days prior), and the measure is reported out quarterly as opposed to monthly for the other measures. A third outcome measure is the mean cJADAS10 score. This measure has similar inclusion and exclusion criteria as our primary measure but assesses cJADAS10 scores from all patients with JIA regardless of subtype.

An additional measure is clinical inactive disease by the American College of Rheumatology (ACR) provisional criteria with the exclusion of inflammatory markers (41). The patient needs to fulfill all five criteria at their last visit to be included in the measure: (1) active joint count of zero, (2) no systemic features (only applicable if a patient has systemic JIA), (3) physician global assessment of disease activity of zero, (4) morning stiffness of 15 min or less, and (5) no current active uveitis. All JIA patients with at least two clinic visits in the past 450 days, with the second visit being at least 180 days after their diagnosis, are eligible for this measure. The patient is excluded if any of the ACR provisional criteria are missing. Additional outcome measures related to disease activity include the mean active joint count and the mean physician global assessment of disease activity score. These measures include all patients with JIA and have similar inclusion and exclusion criteria as our primary measure.

Analysis

Data are collected at member sites by manual chart review and abstraction and/or electronic data transfer between the EHR and the PR-COIN registry. Site data are pooled to populate collaborative measure data, and this is displayed over time on run charts or control charts. Data span from 2011, when the network

was created, to March 2024. Initial center lines are calculated from the initial 20 data points. Special cause on control charts was determined by the presence of two standard control chart rules: (1) shift – 8 or more points in a row above or below the center line and (2) trend – 6 consecutive points increasing or decreasing (42). Furthermore, for run charts, the following standard rules were utilized to determine special cause: (1) shift – 6 or more points in a row above or below the center line and (2) trend – 5 consecutive points increasing or decreasing (42).

Ethical considerations

The PR-COIN registry protocol was approved by Seattle Children's Institutional Review Board (IRB), which serves as the IRB of record for Seattle Children's Hospital and the following relying participating sites: Stanford University, University of Mississippi, Children's Wisconsin, Northwell Health/Cohen Children's Medical Center, Baylor College of Medicine/Texas Children's Hospital, University of Minnesota, Phoenix Children's Hospital, Nationwide Children's Hospital, Medical University of South Carolina, Hospital for Special Surgery, Hackensack Meridian Health, Cincinnati Children's Hospital Medical Center, Children's Mercy Kansas City, Children's Hospital of Philadelphia, Boston Children's Hospital, and University of Alabama at Birmingham. Due to institutional regulatory policies and local or provincial laws and regulations, the PR-COIN registry protocol was approved by a local IRB for the following participating sites: Levine Children's/Atrium Health (Charlotte, NC, United States), London Health Sciences Centre/Lawson Health Research Institute (London, ON, Canada), McMaster University (Hamilton, ON, Canada), Nemours Orlando (Orlando, FL, United States), Penn State Children's Hospital (Hershey, PA, United States), and The Hospital for Sick Children/SickKids (Toronto, ON, Canada).

Results

Five outcome measures related to disease activity have shown improvement over time. The inactive or low disease activity by cJADAS10 measure (Figure 2) has significantly improved over the last several years with shifts in the data. The initial mean in 2012 and 2013 was 71%, and the current center line is at 81%. The performance for clinical inactive disease by ACR provisional criteria (Figure 3) started at a mean of 46%. After upward shifts, the average collaborative performance is now 60%. The mean disease activity by cJADAS10 measure (Figure 4A) has improved from 4.3 to 2.6 after numerous shifts in the data. In 2011 to mid-2013, the mean active joint count (Figure 4B) was 1.5. This number has significantly decreased over the years with the current center line indicating a mean active joint count of 0.7. Mean physician global assessment of disease activity score (Figure 4C) also significantly improved from 1 to 0.6. The final disease activity outcome measure, inactive or low disease activity by cJADAS10 by six months after diagnosis, has not shown any significant improvement. Quarterly performance from January 2022 to March 2024 has ranged from 30% to 80%.

Discussion

Summary

PR-COIN has made significant improvements in the network's disease activity outcome measures for patients with JIA.

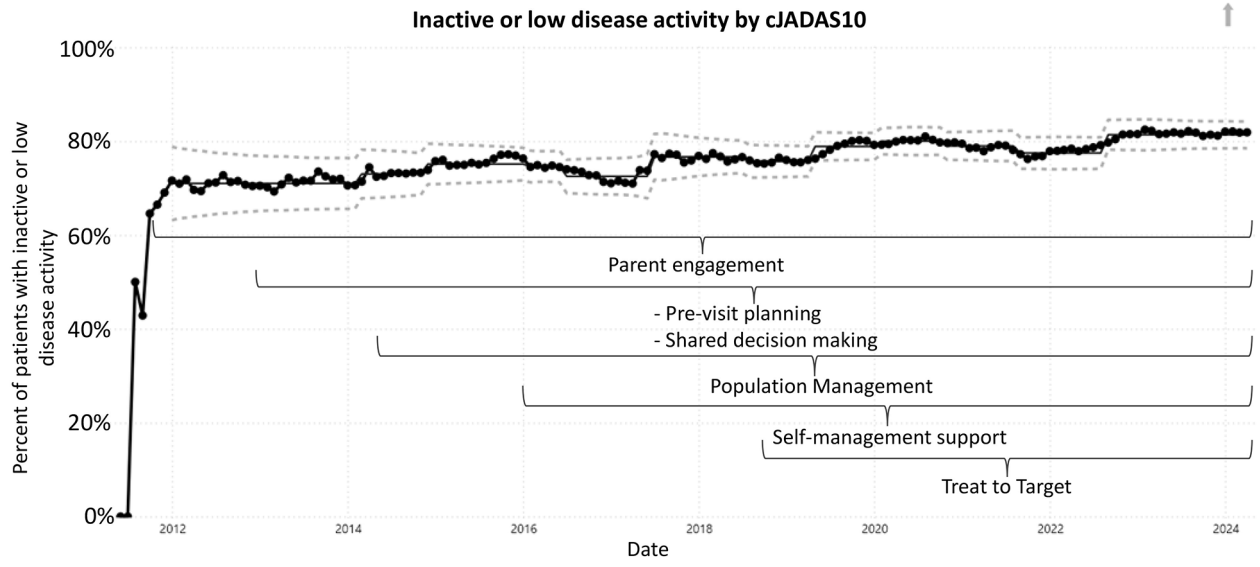

FIGURE 2
Control chart assessing inactive or low disease activity by cJADAS10. The dots represent our monthly performance. The center line is the mean, and the dashed lines are the upper and lower control limits.

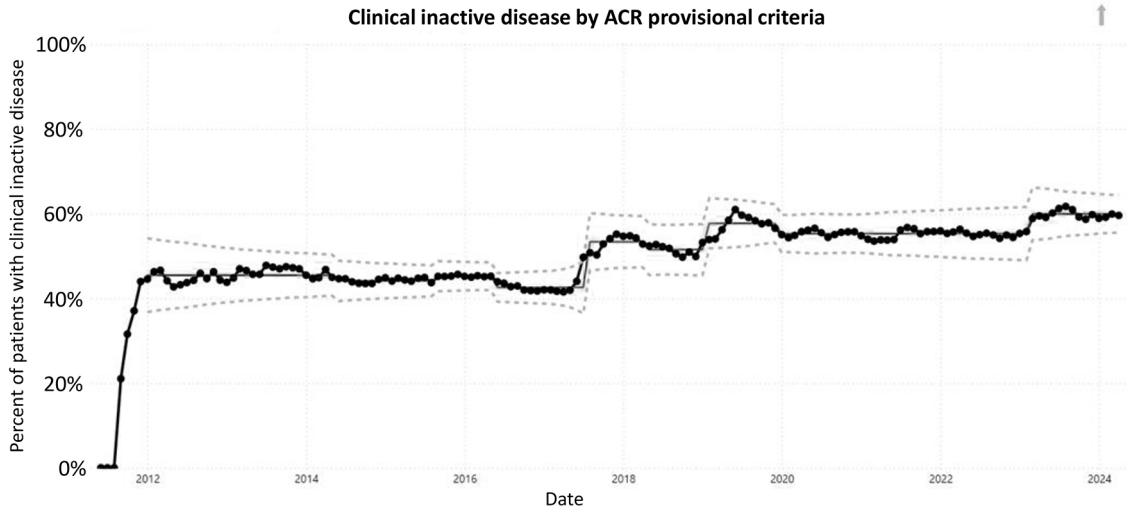

FIGURE 3
Control chart assessing clinical inactive disease per ACR provisional criteria. The dots represent our monthly performance. The center line is the mean, and the dashed lines are the upper and lower control limits.

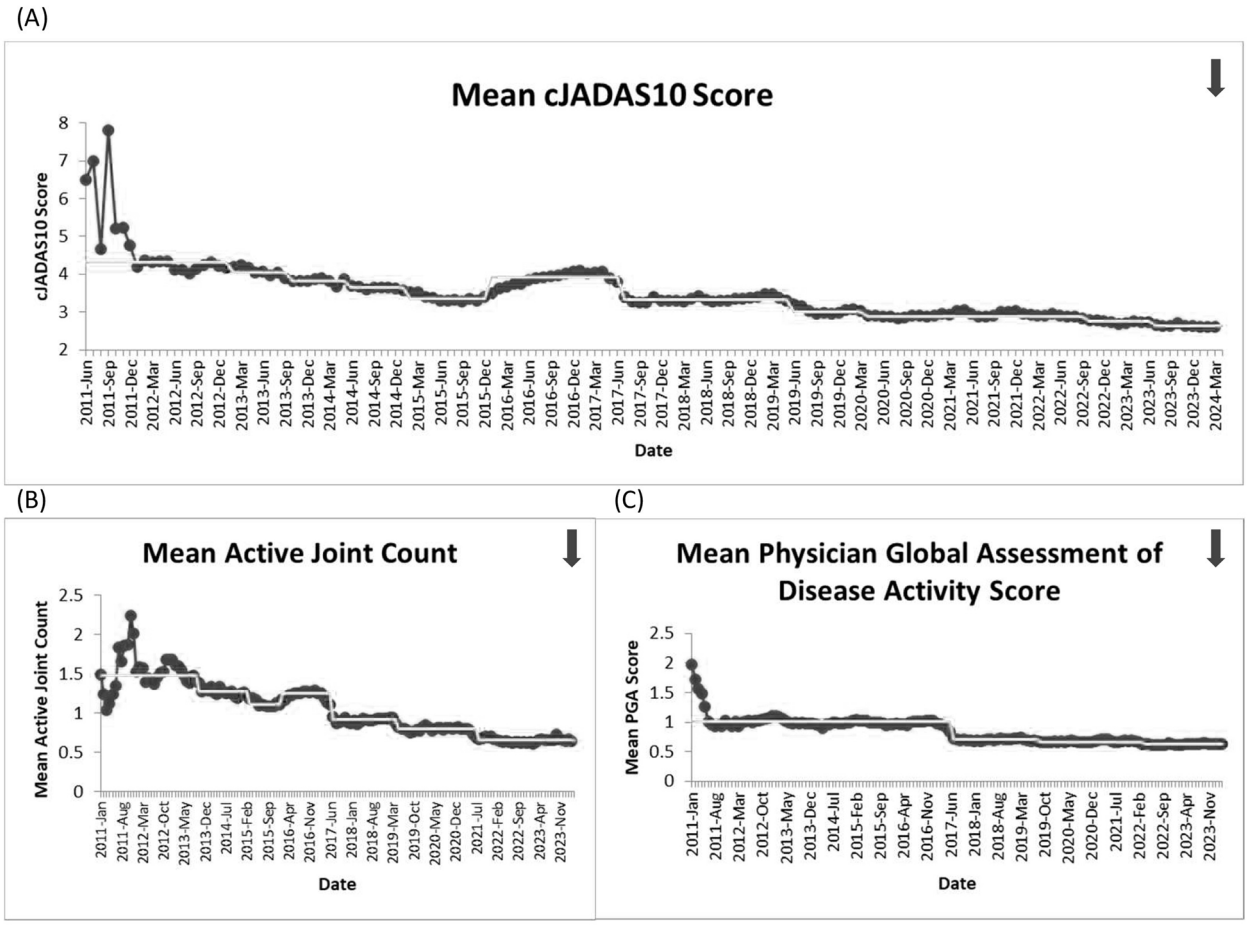

FIGURE 4
Run charts of the following measures: (**A**) mean disease activity by cJADAS10, (**B**) mean active joint count, and (**C**) mean physician global assessment of disease activity score. The dots represent our monthly performance, and the center line is the median.

Furthermore, the network surpassed its goal to have 80% of patients with oligoarticular or polyarticular JIA in an inactive or low disease activity state measured by cJADAS10. Positive substantial change was also noted to the mean cJADAS10 score, the mean active joint count, the mean physician global assessment of disease activity score, and the percent of patients with JIA in clinical inactive disease per ACR provisional criteria.

Overall, PR-COIN's QI initiatives play a vital role in driving continuous improvement in outcome measures for JIA by fostering a culture of learning, collaboration, and innovation within the pediatric rheumatology community. This is the first manuscript highlighting performance on outcome QMs for JIA over time in a quality improvement learning network. PR-COIN's structure and focus on transparency and sharing of best practices has contributed to these improvements in addition to the use of QI methodology both at sites and as a network. PR-COIN's membership across numerous academic pediatric centers throughout the United States and Canada allowed for thousands of patients to be included in these measures, making the results even more meaningful. Numerous network-supported interventions contributed to these improvements including pre-visit planning, shared decision making, self-management support, population management, and a Treat to Target approach to care. Involvement of patients and families in a co-production model has also positively contributed to the network's improvements.

Interpretation

Direct comparison of our outcome measure performance to other JIA populations in the literature is challenging, and there is a paucity of studies in the literature looking at performance on these validated outcome measures over time. An older Canadian cohort of 16 centers analyzed disease activity outcomes in their combined JIA population; however, direct comparisons to our data is challenging given their different outcome measure definitions and timing of evaluating these outcomes being based off of disease duration (43). This study noted that more than 70% of patients with JIA were in inactive disease within 2 years of diagnosis for all JIA subtypes except rheumatoid factor positive polyarticular JIA patients. Hissink Muller et al. noted that 71% of recent-onset patients with JIA had inactive disease following a 24-month period of providing treat to target-based care (44). Patients with JIA from the Childhood Arthritis and Rheumatology Research Alliance (CARRA) Registry were evaluated at one point in time at least 1 year from diagnosis (45). Forty-six percent of this population had clinical inactive disease by ACR provisional criteria compared to our 60% of patients with JIA. The cJADAS10 measure they reviewed looked at patients with score of 1 or less, which differed from our definition assessing for inactive or low disease activity by established cut-offs by subtype (40). This study's cohort had a median cJADAS10 score of 2 in comparison to our final mean cJADAS10 score of 2.6 (45). Additionally, the authors from the referenced CARRA Registry study noted 51% of patients had a physician global assessment of disease activity score of 0 with median of 0; our last center line of the mean physician global assessment of disease activity score was 0.6.

Limitations

Our work has some limitations related to data quality. Some sites contribute a relatively small number of patients to each measure given potential local factors including provider engagement, data collection practices, and the time-consuming process of manual data entry. Representativeness of data is lacking in regard to newly diagnosed patients with JIA. This likely contributed to our variability in performance and lack of improvement in our outcome measure of inactive or low disease activity by cJADAS10 by 6 months (after diagnosis). Although 23 sites are participating in the PR-COIN network now, data are actively being entered on a regular basis by 15–17 sites. Transitions between registry platforms have also led to occasional interruptions in data transmission as well.

PR-COIN tracks outcomes for all patients with JIA that can be enrolled in the registry from participating pediatric rheumatology centers. These patients have varying backgrounds and disease severities, which is a strength of this type of analysis, as real-world practice is reflected. This is one reason why data completeness and timeliness are emphasized so that the registry can be representative of all patient populations. There have been teams that have joined and left the network over the past several years that could have influenced QM performance, although they contributed a small number of patients, and the impact is likely minimal. In addition, due to patients aging out of pediatrics, the active patient population changes over time so this is not the same group of patients from year to year.

As highlighted in the Methods section, PR-COIN participates in several network-level interventions or interventional themes. Most of these have overlapping times of initiation/adoption and continued engagement on, which can lead to uncertainty into what interventions directly lead to improvements. Sites also may be working on their own QI projects related to JIA, and the network does not systematically track these individual projects over time. PR-COIN plans to annotate network charts more and encourage sites to track their projects/interventions as well to determine if a change is temporally related. It is possible that changing medication and treatment practices have occurred during the study timeframe that may have partially accounted for a secular trend towards outcome improvement over time.

PR-COIN had a network aim for one of its outcome measures, the inactive or low disease activity by cJADAS10 measure. However, there were no set goals for the other disease activity outcome measures. PR-COIN is actively setting targets now for all of its QMs, and these goals will be reflected on the run charts and control charts going forward. Additional limitations that the network is rectifying include ability to stratify our outcome measure data by numerous variables including race, ethnicity, age, sex assigned at birth, JIA subtype, disease duration, insurance status, and more. Future direction includes ability to consider patient mix when comparing performance across

centers. For example, centers with more severe phenotypes (polyarticular, rheumatoid factor positive) unadjusted may show lower rates of disease control compared to other centers. Although the focus of this manuscript was on the disease activity outcome measures, it is important to note that PR-COIN has several patient-reported outcomes that were outside the scope of this manuscript.

Conclusions

PR-COIN has demonstrated significant improvements in disease activity outcomes for patients with JIA over time. With continued use of QI methodology for both site-specific and collaborative projects, PR-COIN will continue to live out its mission of using QI science to deliver exceptional and equitable health care to children with JIA.

Data availability statement

The datasets presented in this article are not readily available due to legal and IRB requirements. Requests to access the datasets should be directed to PR-COIN@seattlechildrens.org.

Ethics statement

The studies involving humans were approved by the Seattle Children's Institutional Review Board (IRB), which serves as the IRB of Record for Seattle Children's Hospital and the following relying participating sites: Stanford University, University of Mississippi, Children's Wisconsin, Northwell Health/Cohen Children's Medical Center, Baylor College of Medicine/Texas Children's Hospital, University of Minnesota, Phoenix Children's Hospital, Nationwide Children's Hospital, Medical University of South Carolina, Hospital for Special Surgery, Hackensack Meridian Health, Cincinnati Children's Hospital Medical Center, Children's Mercy Kansas City, Children's Hospital of Philadelphia, Boston Children's Hospital, and University of Alabama at Birmingham. Due to institutional regulatory policies and local or provincial laws and regulations, the PR-COIN registry protocol was approved by a local IRB for the following participating sites: Levine Children's/Atrium Health (Charlotte, NC, United States), London Health Sciences Centre/Lawson Health Research Institute (London, ON, Canada), McMaster University (Hamilton, ON, Canada), Nemours Orlando (Orlando, FL, United States), Penn State Children's Hospital (Hershey, PA, United States), and The Hospital for Sick Children/SickKids (Toronto, ON, Canada). The studies were conducted in accordance with the local legislation and institutional requirements. The Ethics Committee/institutional review board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because the research involves no more than minimal risk to research subjects and their privacy. In addition, the waiver of consent, assent and/or HIPAA authorization will not adversely affect the rights, including the right to privacy, nor adversely affect the welfare of the subjects whose records or data are being used. Furthermore, the research could not practicably be carried out without the waiver of consent and/or HIPAA authorization. Of note, some centers do require written informed consent per local IRB requirements.

Author contributions

JH: Conceptualization, Data curation, Methodology, Writing – original draft, Writing – review & editing. CB: Conceptualization, Data curation, Methodology, Writing – original draft, Writing – review & editing. SV: Data curation, Methodology, Writing – original draft, Writing – review & editing. CY-T: Methodology, Writing – original draft, Writing – review & editing. MB: Data curation, Writing – review & editing. DB: Methodology, Writing – review & editing. JB: Data curation, Methodology, Writing – review & editing. DF: Data curation, Writing – review & editing. KF: Methodology, Writing – review & editing. SG: Data curation, Writing – review & editing. MG: Data curation, Writing – review & editing. BG: Data curation, Writing – review & editing. OH: Writing – review & editing. MH: Writing – review & editing. RL: Data curation, Writing – review & editing. TL: Writing – review & editing, Data curation. AL: Writing – review & editing, Project administration, Writing – original draft. DL: Data curation, Writing – review & editing. MM: Data curation, Writing – review & editing. EO: Data curation, Writing – review & editing. NP: Data curation, Writing – review & editing. MS: Data curation, Writing – review & editing. JW: Data curation, Writing – review & editing. EM: Conceptualization, Data curation, Methodology, Project administration, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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The critical role of parents within a Learning Health Network

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Parent members of the Pediatric Rheumatology Care & Outcomes Improvement Network are an integral part of the Learning Health Network's work. Since early in the creation of the network, they have been a part of every Quality Improvement project, committee, and work group and have a role in governance on the Executive and Steering Committees. Members of the Parent Working Group (PWG) have played a role in developing QI measures used in the clinical setting as well as initiatives and projects like the guiding work of Treat-to-Target. The PWG also creates self-management supports, including toolkits for families and patients at all stages of life. This article will discuss how integrating parents as partners in a pediatric Learning Health Network is critical for the quality of care received by children with chronic illnesses and to improving outcomes.

KEYWORDS

engagement, parent, partners, juvenile arthritis, quality improvement, pediatric rheumatology, Learning Health Network, co-production

Introduction

Parent members of the Pediatric Rheumatology Care & Outcomes Improvement Network (PR-COIN) have been an integral part of the Learning Health Network since soon after its formation in 2010. As early as 2012, PR-COIN invested resources to build capacity for engaging families at its centers. A Parent Engagement Consultant was hired to create and accelerate strategies for participation and engagement of families at both individual member team and PR-COIN network levels. In 2014, parents were invited to join Committees, including the Steering Committee, and funding was provided for three parents to attend the Learning Session.

The role of the parents took off in 2015 when the parent group chose to name itself the Parent Working Group (PWG) and to better define their mission and objectives within PR-COIN. PR-COIN added family engagement to its center agreements. Parents co-presented with providers at the Learning Sessions, and created toolkits and resources. The Engagement Committee was created, with this article's co-author Kerry Ferraro being named as the lead. She also became the first parent to join the Executive Committee in 2019. Since then, PR-COIN has enshrined patients with Juvenile Idiopathic Arthritis (JIA) and their families in its approach and its mission "to build a thriving and inclusive community of patients, families, clinical teams and researchers that uses quality improvement (QI) science to deliver exceptional and equitable health

care to children with rheumatic diseases and to bring research discovery to patients faster." This approach involves including patients and families in all levels of governance and network activities to ensure patient-centered care. Parents are partners in the work (1).

PR-COIN's Key Driver Diagram, shown in Figure 1, identifies Engaged Patients and Families as a crucial key driver. Several of the changes and interventions explicitly emphasize parent engagement, such as Embedded parent/patient member; Connect to Parent/Patient Working Groups; Facilitate patient/family input/development/improvement of care delivery; and Co-production between all stakeholders. Moreover, parents have significantly contributed to the implementation of other changes and interventions outlined in the Key Driver Diagram.

A key member of PR-COIN's leadership team, Dr. Julia Harris wrote in her paper, "Improving Care Delivery and Outcomes in Pediatric Rheumatic Diseases" (2): "Engaging patients and families is an increasingly recognized component for quality improvement in healthcare that is patient centered. PR-COIN has formed a parent leadership roundtable, Facebook page, parent attendance at action period calls, and robust parent presence at learning sessions. Parents set 90-day goals, hold positions at leadership level and on PR-COIN subcommittees, and contribute novel talent and skills to the network. Through patient/parent engagement, PR-COIN is striving to foster co-production of network activities whereby health care teams and patients/families can work together to produce more desirable outcomes for children with JIA."

Beyond the governance and co-production areas, parent support extends to the organization's fiscal health. Parents are key members of the Finance and External Partnerships Committee and have written letters to hospital leadership to encourage them to join or remain a member of PR-COIN. They have been involved in the creation of marketing and fundraising materials, held fundraisers, and assisted in securing grants. This not only supports the organization, but it also eases the financial burden on the centers.

Parents and families are an integral part of the guiding work and ability of PR-COIN to achieve QI. This article will discuss the critical role of parents in implementing the changes and interventions that support the key drivers of improvement within a pediatric Learning Health Network by exploring examples of parent involvement in four specific interventions under the categories of Robust QI Capability and Capacity, Equipped Families, Co-Produced Care, and Robust, Reliable Care. In this article, "parent" is being used to refer to the primary caregiver.

Key driver diagram

PR-COIN's Key Driver Diagram (KDD), shown in Figure 1, identifies the network's AIMS as of December 31, 2023 and highlights the key drivers of Access to care; Timely diagnosis and treatment per individual targets; Prepared, proactive practice team; Engaged Patients and Families; and Culture facilitating QI and research. The changes and intervention categories on the

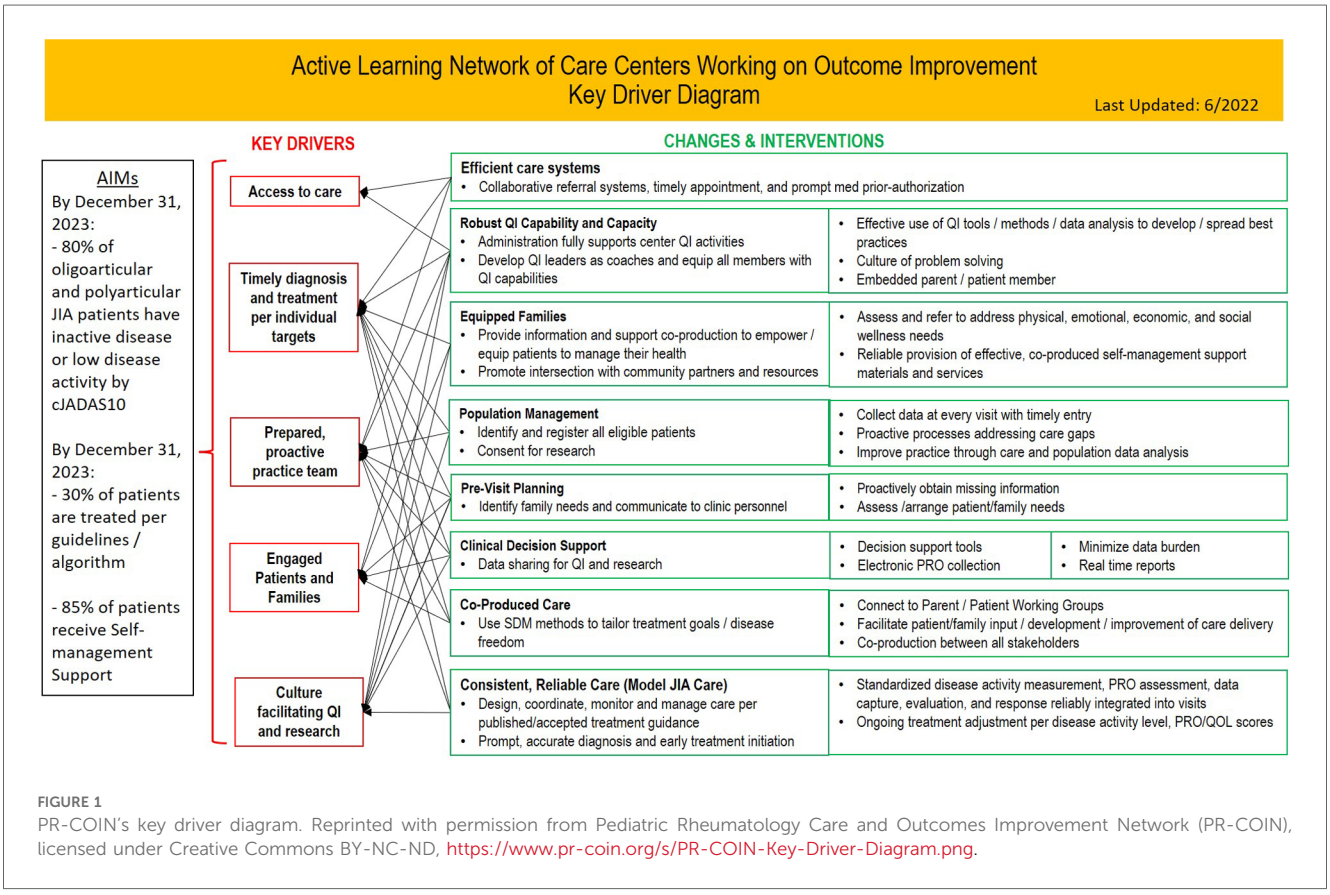

FIGURE 1
PR-COIN's key driver diagram. Reprinted with permission from Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN), licensed under Creative Commons BY-NC-ND, https://www.pr-coin.org/s/PR-COIN-Key-Driver-Diagram.png.

right of the KDD are Efficient care systems; Robust QI Capability and Capacity; Equipped Families; Population Management; Pre-Visit Planning; Clinical Decision Support; Co-Produced Care; and Consistent, Reliable Care (Model JIA Care). We will explore specific interventions that fall under four of these categories.

Robust QI capability and capacity

Within the Robust QI Capability and Capacity category, the intervention "Embedded parent/patient members" explicitly emphasizes parent engagement. Additionally, voices of parents are heard at the very core of PR-COIN's work—the development of the quality improvement measures used to track patient outcomes under the Effective use of QI tools intervention. As shown on the KDD, these interventions support each key driver of the network.

Dr. Catherine Bingham's paper "Pediatric Rheumatology Care and Outcomes Improvement Network's Quality Measure Set to Improve Care of Children With Juvenile Idiopathic Arthritis" (3) notes how each center forms a local QI team and states, "Because the voices of patients and families are invaluable to inform the challenges to care and impact of disease, patient/parent representatives are also included."

A formal PR-COIN Measures Committee was formed in 2013 and was composed of volunteers from center teams and JIA parent representatives. Bingham discussed how the original Quality Measures (QM) were based on a 2008 ACR project to identify QMs for JIA that included surveys of pediatric rheumatologists, APNs, patients, and parents. This was also discussed by Dr. Daniel Lovell et al. in the 2011 paper, "Measuring Process of Arthritis Care" (4).

Parents were also involved in developing the new QM set in 2018 (3). Parents conveyed the importance of including the Patient Global Assessment of overall well-being and emphasized that the provider assessments alone do not capture the lived experiences of youth where, for example, medication reactions might leave a child in bed for days. Therefore, the percentage of patients who had a Patient Global Assessment of overall well-being less than or equal to two was added to the revised QM set by the PR-COIN Measures Committee (3).

Parent and patient voices are especially critical because often there is a discordance between providers and patients in disease assessment, which is the reason parents pushed for the Patient Global Assessment to be included in our QM set (5).

Equipped families

Within the Equipped Families category, parents played a significant role in the interventions of: Provide information and support co-production to empower/equip patients to manage their health and Reliable provision of effective, co-produced self-management support materials and services. These directly support the key drivers of Timely diagnosis and treatment per individual targets and Engaged Patients and Families.

Disease outcomes are impacted heavily by a patient and their parent being informed and able to manage the condition effectively. Self-management activities include managing medications, whether injectable, oral or intravenous, and managing appointments as well as blood work, imaging and home exercise programs. It further extends to the daily life of the youth living with the disease, including going to school, managing health insurance coverage, and managing mental health. Self-management support (SMS) is one of the six essential elements of care identified in the CCM (6). Rheumatology nurse practitioner and former Outcome Committee co-lead, Janalee Taylor championed PR-COIN's SMS initiatives and believes "These activities must be integrated into daily life with consideration for developmental, intellectual, and psychosocial well-being of the child and family" (7).

Parents were included in the work groups for each of the tools created as part of the SMS system, which included the Barriers Assessment, Adherence Tools, Self-Management Assessment, and Helping Hands Handbook. Parents also co-presented on the tools at a Learning Session and encouraged providers to use the tools.

Member centers are provided with a SMS System Change Package. This is a toolkit to support practitioners and families in formulating and adhering to medication and treatment plans. The change package includes provider training in Behavior Change Counseling, and use of Self-Management Assessment Tool, Barriers Checklist, Adherence Solution Tools, Patient Action Plan worksheet, and complete family educational materials in the Helping Hands Handbook (8).

The Self-Management Assessment identifies patient/family priorities for visit, level of confidence in ability to manage disease, level of worry, goal setting, adherence and general barriers to care. The Barriers Checklist helps providers identify what comes in the way of following treatment. For each barrier, an adherence solution is identified, including corresponding tools and templates. The Helping Hands Handbook contains information about JIA, medications, treatments, and activities to optimize quality of life.

The Barriers Assessment provides a good example of how these toolkits are developed (9). Providers and parents worked together through an iterative process to design the Barriers Assessment Tool to screen for adherence barriers across 4 treatment modalities (i.e., oral medications, injectable medications, infusions, and physical/occupational therapy). This tool was initially implemented in seven rheumatology clinics across the United States and patient responses were collected for analysis. Seventy-seven percent ($n = 444$) of caregivers and 70% ($n = 69$) of patients reported at least one adherence barrier (9). Identifying the barriers enabled care teams to provide the tools needed to overcome the barriers to adherence.

Equipping families expands beyond the tools created as part of the SMS tools. The PWG identifies and creates tools and resources to assist patients and their families in navigating life with JIA. Navigating school with JIA is often a challenge, so we adapted ICN's Accommodation Toolkit, which provides information on ADA laws and academic, workplace, and public accommodations

for patients with JIA. Five parents helped, but parent Laura Bouslaugh's expertise as a Civil Rights Specialist was especially helpful. Next, parents teamed up with young adult patients and created a College and JIA toolkit, which provides information on finding a "good fit," preparing to go to college with JIA, tips for being successful in college with JIA including accommodations and how to request them, taking time off college, and personal perspectives of JIA patients.

This process is iterative. We learned from center feedback that the Helping Hands Handbook was not being provided to patients at all centers, so we created a one-page frequently asked questions resource that includes a QR code to the handbook. This allows centers to only print a single page rather than a 127-page handbook. We also created a one-page accommodations summary that links to the 23-page accommodations toolkit. Next, we created the insurance toolkit, which helps families navigate health plans, including prior authorizations, step therapy, and denied claims. We surveyed parents to identify what educational materials were needed at the time of diagnosis to identify gaps. One third of the respondents wrote in that they wanted information on ways to connect with other patients and families. We created a resource list for families that includes links to the PR-COIN toolkits, Arthritis Foundation resources, support groups, social media resources, and podcasts.

Co-produced care

Co-producing care calls for including parents in the co-production of the care and the improvement work of the network under the interventions of: Connect to Parent/Patient Working Groups; Facilitate patient/family input, development and improvement of care delivery; and Co-Production between all stakeholders. It also calls for the Use of SDM (shared decision making) methods to tailor treatment goals and disease freedom.

PR-COIN follows the Chronic Care Model which states that quality care involves productive interactions between informed, activated patients and their care teams (6). To improve these interactions, one of PR-COIN's early interventions was the use of SDM methods. PR-COIN's Medication Choice Cards help improve parent and patient engagement and facilitate the discussions required for SDM.

Parents played a role in the development and revisions of Medication Choice Cards, shown in Figure 2. These cards walk parents and patients through any choices they may be facing, allowing them to be informed and involved. The initial design of the cards involved an iterative process with a stakeholder panel of parents and care team members (10). Providers and parents co-presented the use of the Medication Choice Cards at a PR-COIN Learning Session. Six sites volunteered to use QI

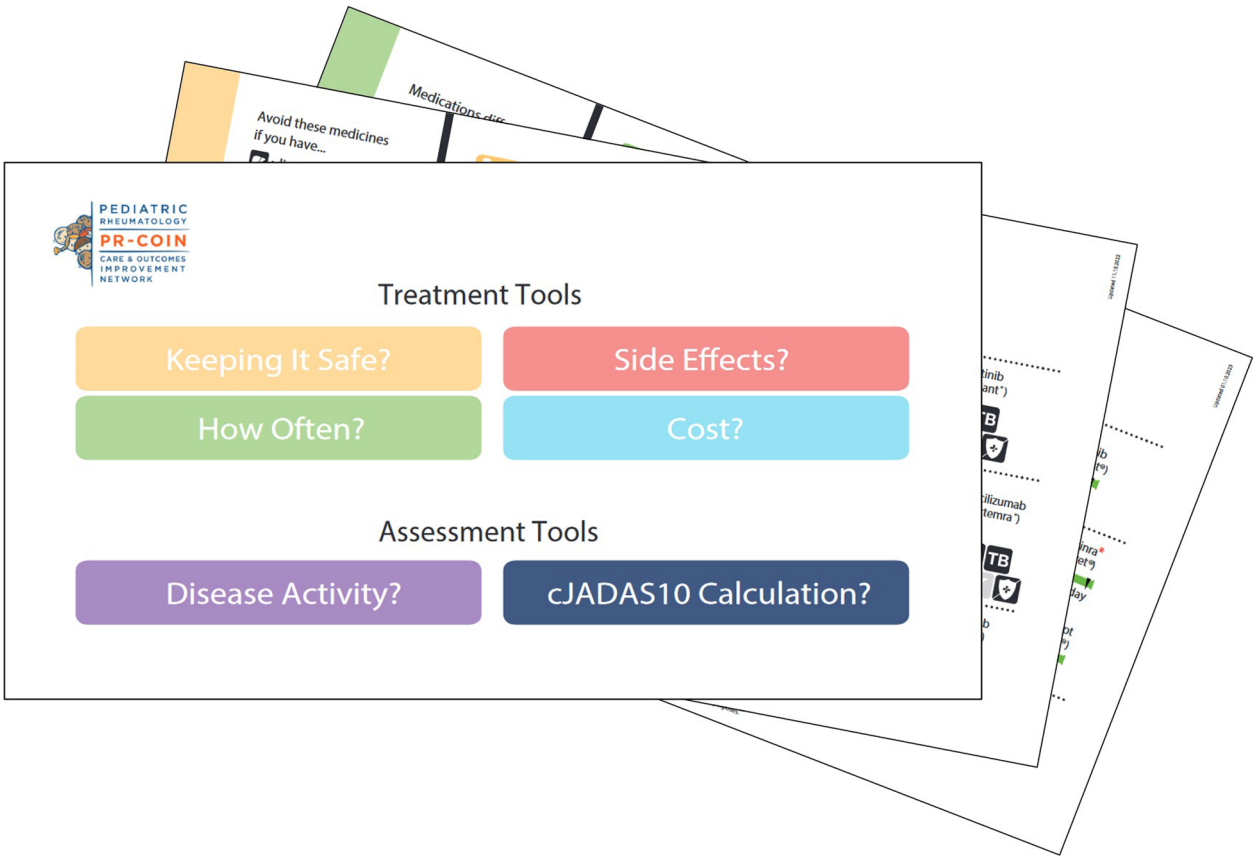

FIGURE 2
Sample image of PR-COIN's medication choice cards.

methods to implement the cards. Four of these sites collected parent surveys following visits to assess outcomes. Patients and parents shared on clinician use of the cards and the amount of SDM and uncertainty they experienced (11). The study authors concluded that more reliable use and sharing of best implementation practices was needed and questioned whether an electronic format may lead to more reliable use. In response, Ferraro created an interactive, digital format of the cards for use by clinicians. Another case study showed that use of the "Medication Choice Cards significantly enhanced shared decision making in treatment of adolescents with JIA, through increased patient engagement" (12).

As treatment options change, parents continue to partner in the updates and revisions to the paper and digital cards as well as to the pamphlet that was developed for parents to make notes on and take home to discuss the treatment choices with other family members.

Consistent, reliable care

The key drivers include: Timely diagnosis and treatment per individual targets; Prepared, proactive practice team, and Engaged patients and families. To support these, PR-COIN identified the need for Consistent, Reliable Care as an intervention category. Due to its focus on all of the interventions under the Consistent, Reliable Care category on the KDD in Figure 1, Treat-to-Target (T2T) has become the foundational work of PR-COIN.

Dr. Sandy Burnham started T2T with polyarticular JIA and included a polyarthritis parent in the project meetings. Work on T2T began in 2004 when it was noted that not enough patients were achieving inactive disease. T2T takes the core concepts of measure, standardize, and use of clinical decision support and applies them in the clinical setting. As Burnham developed the project, he shared the patient-facing materials with parents for feedback. When PR-COIN decided to make the project network-wide, parents were partners in creating the educational materials for providers and families. "Designing and Testing Treat to Target as a New Care Model in JIA Across a Network of Pediatric Rheumatology Centers" notes that "With patient/family partners, PR-COIN co-produced educational materials to train providers on implementation of T2T and to introduce families to the concept…. Co-producing support materials with families, infrastructure to support QI, and reliable data submission are key to success" (12).

In 2020, PR-COIN held a consensus conference to standardize treatment plans (14). "PR-COIN stakeholders, including health care providers ($n = 16$) and parents ($n = 4$), were invited to form a voting panel." When identifying the most important elements when setting an individual's target, parents advocated for patient goals. After the second round of voting, "patient goals" was voted unanimously as the most important element. Parents revised the Medication Choice Cards to include the clinical Juvenile Arthritis Disease Activity Score (cJADAS) calculation and disease activity level by JIA subtype to help identify if the disease level is at target. Since the cJADAS includes both the Physician Global Assessment and the Patient Global Assessment of wellbeing, this gives the clinicians the opportunity to discuss any discordance between the clinician's assessment of disease activity and the parent's or patient's assessment.

All these tools create a clinical practice that involves parents. In one example provided by a provider, an 18-month-old patient came to her with polyarticular JIA and very sick. The PR-COIN method of approaching the mom from her perspective and using tools like shared decision making and T2T intervention helped the mom gain more acceptance of the treatment. Her anxiety level came down as the child was doing better and as she understood options and saw that labs helped monitor safety (15).

Discussion

Beginning with early investments in parent engagement and the establishment of the Parent Working Group, PR-COIN has recognized and integrated the invaluable insights of parents into its governance and improvement initiatives. This collaborative approach has enriched the network's strategies and has empowered families.

The involvement of parents in pivotal roles such as committee membership, co-presentation at learning sessions, and co-production of essential tools and interventions underscores the impact of parents on care delivery and patient outcomes. Including parents as partners in a pediatric learning health network (LHN) fosters a deeper level of engagement with parents and patients during clinic visits and leads to a more patient-centered approach. PR-COIN's commitment to patient-centered care is evident in how parents and patients are integrated at every level of Key Drivers and Changes and Interventions. By championing shared decision making and advocating for patient goals in treatment planning, parents have fostered a culture of trust and collaboration between healthcare providers and families.

The impact of parent involvement in PR-COIN extends beyond clinical settings, influencing network governance, financial sustainability, and outreach efforts aimed at expanding PR-COIN's impact. PR-COIN parents are currently leading the development of an engagement measure to better evaluate how centers engage parents in their work, aiming to quantitatively assess the level of engagement and its impact outcomes of QI initiatives.

Looking ahead, future studies utilizing this engagement measure could provide insight on the correlation between parent engagement and improved health outcomes within pediatric LHN. Other LHNs who wish to improve parent and patient involvement in healthcare decision-making and network governance would do well to look at PR-COIN as a model.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material, further inquiries can be directed to the corresponding author.

Author contributions

KF: Writing – original draft, Writing – review & editing. JL: Writing – original draft, Writing – review & editing. AS: Writing – original draft, Writing – review & editing. SP: Writing – original draft, Writing – review & editing. CP: Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Electronic health record modification and dashboard development to improve clinical care in pediatric rheumatology

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Objective: This report describes our experience in electronic health record (EHR) note modification and creation of an external dashboard to create a local learning health system that contributes to quality improvement and patient care within our pediatric rheumatology clinic.
Methods: We applied quality improvement methodology to develop a more reliable and accurate system to identify patients with juvenile idiopathic arthritis and track important measures that aide in improving patient care and performance outcomes. From 2019 to 2021, we iteratively modified our outpatient clinic EHR note to include structured data elements to improve longitudinal monitoring. We then validated data transferred to an electronic dashboard external to the EHR and demonstrated utility for identifying an accurate patient population and tracking quality improvement initiatives.
Results: Creation of the structured data elements improved the identification of patients with JIA with >99% accuracy and without requiring manual review of the chart. Using the dashboard to monitor performance, we improved documentation of critical disease activity measures that resulted in improvement in those scores across the local population of patients with JIA. The structured data elements also enabled us to automate electronic data transfer to a multicenter learning network registry.
Conclusion: The structured data element modifications made to our outpatient EHR note populate a local dashboard that allows real time access to critical information for patient care, population management, and improvement in quality metrics. The collection and monitoring of structured data can be scaled to other quality improvement initiatives in our clinic and shared with other centers.

KEYWORDS

electronic health record, dashboard, population management, juvenile idiopathic arthritis, pediatric rheumatology

Introduction

Technology advancement and universal use of electronic health records (EHR) has allowed providers new ways to collect and track quality measures and improvements within healthcare. While the purpose of the health record is to document medical care, an electronic record can be leveraged to capture specific aspects of care and serve as a tool to efficiently access and analyze care processes, specific disease measures, and health outcomes (1, 2). These quality measures serve as benchmarks for evaluating the

effectiveness, safety, and efficiency of health care services, facilitating the monitoring and improvement of clinical practices and patient outcomes (3).

Routine measurement and monitoring of clinical disease activity and care processes is especially important for patients with chronic diseases and is facilitated by an easy-to-use system (4). The most common type of chronic arthritis among children is juvenile idiopathic arthritis (JIA), an autoimmune disease affecting approximately 1 in 1,000 children (5). Individuals with JIA require longitudinal treatment to reduce complications of inflammation and have frequent healthcare interactions for evaluation of disease activity, medication toxicity monitoring, and screening for extraarticular manifestations like asymptomatic uveitis (6–9). Pediatric rheumatologists use various clinical measures to assess disease activity, treatment efficacy, and quality of life (10). These measures have been utilized in studies of a patient-facing dashboard to facilitate patient education and shared decision-making in pediatric rheumatology studies (11, 12).

Clinical outcomes and quality measure performance can be monitored in a clinic population or across clinical sites within the infrastructure of a learning network (13). For example, the Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) is a pediatric rheumatology specific quality improvement learning network that has a centralized patient registry with a dashboard to display network and site-specific processes and disease activity outcomes for patients with JIA (14, 15). Participating sites contribute patient-specific data to the centralized registry with the goal of capturing every JIA patient at every visit to allow for population management (14), a process that has been facilitated by the use of electronic data capture from the EHR with automatic uploading to the registry.

The goal of this initiative was to create an efficient and accurate process for identifying patients with JIA in our EHR, access and track key metrics relate to patient outcomes and clinical care decisions for patients with JIA and automate structured data transfer to the PR-COIN Registry. With the advisement and collaboration of our Information Technology (IT) department, we were able to modify our EHR documentation, create an external dashboard using EHR data that updates in real time, and utilize electronic data transfer to contribute data from our local population to a centralized multicenter registry. We detail our experience with iterative note modifications to create structured data elements within the EHR, utilizing a clinic dashboard for monitoring quality metrics in our population of patients with JIA, and automating data transfer to a multicenter learning network registry.

Methods

Context

Children's of Alabama (COA) is a tertiary care children's hospital in conjunction with the University of Alabama at Birmingham (UAB) that provides comprehensive specialty and subspecialty care for the children of Alabama. Our COA/UAB pediatric rheumatology clinic serves approximately 225 patients each month with varying rheumatic conditions. The outpatient clinics at COA were transitioned to an Allscripts-based EHR in September 2017, and specialty-specific note customization began in 2019 (Figure 1). Our clinical care team at the time of this initiative consisted of 5 attending physicians, 2 clinical pediatric rheumatology fellows, 4 nurse practitioners, and 5 registered nurses. Our clinic is also supported by 4 administrative staff, a medical social worker, and a dedicated research coordinator. Prior to this initiative, there was not a standardized method of identifying patients with JIA so identification of patients for research and abstraction of disease activity measures were performed manually by the providers or research coordinator.

EHR modification and patient identification

After transition to the new EHR system, we received weekly reports from IT that identified patients with JIA by International Classification of Diseases- 10th edition (ICD-10) codes. However, these data requests then required manual review for completeness and accuracy. At the time of the outpatient EHR launch in September 2017, outpatient clinic notes were primarily free text without specialty specific structured data. The rheumatology clinic utilized a specialty-specific note that was developed for inpatient use that included a few clinical data and specific quality measures available in structured fields. For the patients with JIA identified by IT using ICD-10 codes, these data elements (active joint count, physician global assessment of disease activity, patient/parent global assessment of well-being) were provided in the weekly reports emailed by a member of the IT team. However, the measures that were included in the report were often incomplete which limited our ability to monitor and improve care processes and outcomes. Another challenge to analyzing these clinical reports, was that the patients identified by IT with ICD-10 codes resulted in both false positive and false negative results and did not accurately reflect our JIA population. In our clinic the diagnosis codes for billing were collected on paper and the problem lists in the EHR were not required to complete documentation. For each report, every included patient required chart review to confirm a primary diagnosis of JIA and the clinic list for each week would be manually reviewed for possible missed JIA patients.

Beginning in 2019, the rheumatology clinical team and IT collaborated to develop a rheumatology-specific outpatient note that contained structured data elements that were important to clinical care, quality improvement initiatives, and research-based registries (Figure 1). Over the span of two years, we iteratively modified our rheumatology-specific outpatient clinic note to include structured fields (Table 1) for clinical data that would allow for longitudinal monitoring of patients with JIA for quality improvement initiatives and to facilitate data collection for research. We identified key metrics as outlined by the American College of Rheumatology guidelines for treatment of JIA (6, 9, 16, 17), quality metrics identified by PR-COIN (14, 15), and measures important to our clinical team to build data fields

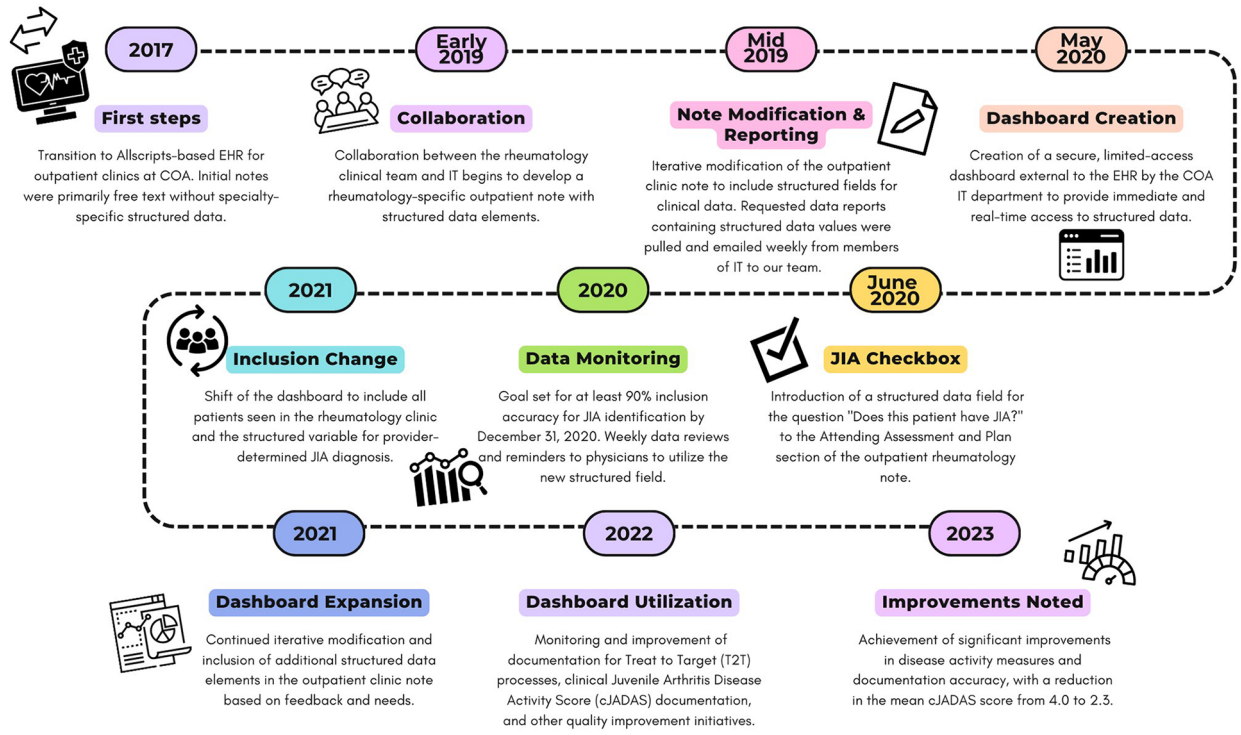

FIGURE 1
Timeline of initiative to improve documentation and review of clinical metrics for JIA in the UAB/COA pediatric rheumatology clinic.

needed to capture these measures from the EHR (Table 1). These measures are collected as part of routine clinical care to determine disease activity and make treatment decisions and include the components and calculated clinical juvenile arthritis disease activity score (cJADAS), calculation of recommended eye screening, and attestation of the components of treat to target (T2 T). The cJADAS is a composite score that includes the physician global assessment of disease activity, the parent/patient assessment of overall well-being, and a count of active joints (maximum of 10). This score ranges from 0 to 30, with higher scores indicating greater disease activity. The goal of T2T is to utilize regular measurement of a standardized assessment and use shared decision making with families to make treatment changes in order to achieve and maintain the lowest possible disease activity to prevent long-term joint damage and improve quality of life (18).

Since relying on ICD-10 code-based definitions led to an inaccurate JIA population, we decided to create a structured field within our note for providers to attest JIA diagnosis. This eliminated the need to manually review query results. In June 2020, we added a structured data field for the question "Does this patient have JIA?" to the Attending Assessment and Plan section of the outpatient rheumatology note (Figure 2). We introduced the new identification process to our providers and set a goal of at least 90% inclusion accuracy by December 31, 2020, giving us approximately 6 months to adjust. Eligible patients included both new and return rheumatology patients with all of the JIA subtypes including oligoarticular, polyarticular (rheumatoid factor positive and negative), enthesitis-related, psoriatic, undifferentiated, and systemic JIA (19). At the visit patients were classified into three categories: JIA yes, JIA no, or unknown. Each selection carried forward to the next clinic note so that the provider only had to mark the appropriate classification once and update if necessary (i.e., if a patient was marked unknown as a new patient and was later determined to have JIA, then the provider would update classification to "JIA yes" at the next visit). Our team utilized various improvement science techniques including statistical run charts and Plan-Do-Study-Act (PDSA) cycles to test changes and track accuracy of the JIA specific checkbox documentation (20). To monitor our progress, we reviewed the data weekly, comparing the patient population identified by ICD-10 codes and the "JIA checkbox" to manual review of the EHR for all patients seen in clinic each week to evaluate the accuracy of patient identification. We tracked the patients in which "JIA yes" should have been checked and sent out weekly reminders to the providers and clinical staff to complete at that patient's next visit. We also put reminder notes on all the computers in the clinic workroom to

TABLE 1 Table of all structured data collected in the UAB/COA pediatric rheumatology clinical outpatient note and dashboard.

Structured data measure	Structure	Available in clinical note	Available on dashboard
All Visits			
Telehealth	Radio	Y	N
Morning stiffness (none, </= 15 min, >15 min)	Radio	Y	N
Inflammatory back pain (Y/N)	Radio	Y	N
Date of last eye exam	Date	Y	N
Last eye exam results (active, past, no uveitis to date)	Radio	Y	N
TB testing date	Date	Y	Y
Hepatitis testing date	Date	Y	N
Home medication	Text	Y	Y
Sum of weight(kg)	Numeric	Y	Y
BMI	Calculated	Y	Y
BP	Numeric	Y	Y
Mouth opening	Numeric	Y	N
Jaw deviation with opening	Radio	Y	N
Notable micrognathia	Radio	Y	N
Modified schober's	Numeric	Y	N
flat back	Radio	Y	N
Scoliosis	Radio	Y	N
Gait (normal/abnormal)	Radio	Y	N
leg length discrepancy (Y/N)	Radio	Y	N
Enthesitis (list for right and left separately: superior patella, inferior patella, Achilles insertion, plantar fascia insertion, metatarsal heads, tibial tuberosity, greater trochanter of femur, elbow condyles)	Radio	Y	N
Left enthesitis count	Calculated	Y	N
Right enthesitis count	Calculated	Y	N
joint assessment (left and right active joint for 72 joints)	Radio	Y	N
Joint assessment (left and right decreased ROM for 72 joints)	Radio	Y	N
Active joint count	Calculated	Y	Y
Decreased ROM count	Calculated	Y	N
transition discussed (Y/N)	Radio	Y	Y
TRAQ score	Numeric	Y	Y
Current glucocorticoid use (Y/N)	Radio	Y	Y
Glucocorticoid type (Oral, IV, other)	Radio	Y	Y
Disease activity assessment (inactive, mild, moderate, severe)	Radio	Y	Y
Pain scale (0–10)	Numeric	Y	N
CHAQ score	Numeric	Y	Y
MD global	Numeric	Y	Y
Parent global	Numeric	Y	Y
cJADAS10 value	Calculated	Y	Y
Treatment target set with family at this visit (Y/N)	Radio	Y	Y
Date target set	Date	Y	N
Target assessment (at target, not at target)	Radio	Y	Y
Disease management change at this visit (Y/N)	Radio	Y	Y
Shared decision-making aid used	Radio	Y	N
Self-management support provided (Y/N)	Radio	Y	N
JIA			
Has JIA ChronicDx (ICD-10 based)	Calculated	N	Y
JIA Yes/No	Radio	Y	Y
JIA subtype (systemic, persistent oligoarticular, extended oligoarticular, oligoarticular unspecified, RF + polyarticular, RF- polyarticular, ERA, psoriatic, undifferentiated)	Drop Down	Y	Y
ANA (+/-)	Radio	Y	N
RF (+/-)	Radio	Y	N
CCP (+/-)	Radio	Y	N
HLA B27 (+/-)	Radio	Y	N
Prognostic features (hip arthritis, wrist arthritis, ankle arthritis, C-spine involvement, radiographic damage, sacroiliitis, TMJ arthritis)	Radio	Y	N
Date of JIA diagnosis	Date	Y	Y
Age at diagnosis (Years)	Calculated	Y	N
Duration of diagnosis (Years)	Calculated	Y	N

(*Continued*)

TABLE 1 Continued

Structured data measure	Structure	Available in clinical note	Available on dashboard
Recommended eye screening interval	Calculated	Y	N
JIA symptoms (present/absent)	Radio	Y	N
Systemic JIA symptoms present (fever, rash, serositis, splenomegaly, generalized lymphadenopathy)	Radio	Y	N
Inflammatory markers (elevated, normal, unknown)	Radio	Y	N
Active uveitis (present, absent, unknown)	Radio	Y	N
Morning stiffness >15 min (Y/N)	Radio	Y	N

TB, *Mycobacterium tuberculosis*; kg, kilograms; BMI, body mass index; BP, blood pressure; ROM, range of motion; TRAQ, transition readiness assessment questionnaire; IV, intravenous; CHAQ, childhood health assessment questionnaire; MD global, physician global disease activity assessment; parent global—parent/patient assessment of well-being; cJADAS10, clinical juvenile arthritis disease activity score 10 joint count; JIA, juvenile idiopathic arthritis; ICD-10, International Classification of Diseases 10th edition; RF, rheumatoid factor; ERA, enthesitis related arthritis.

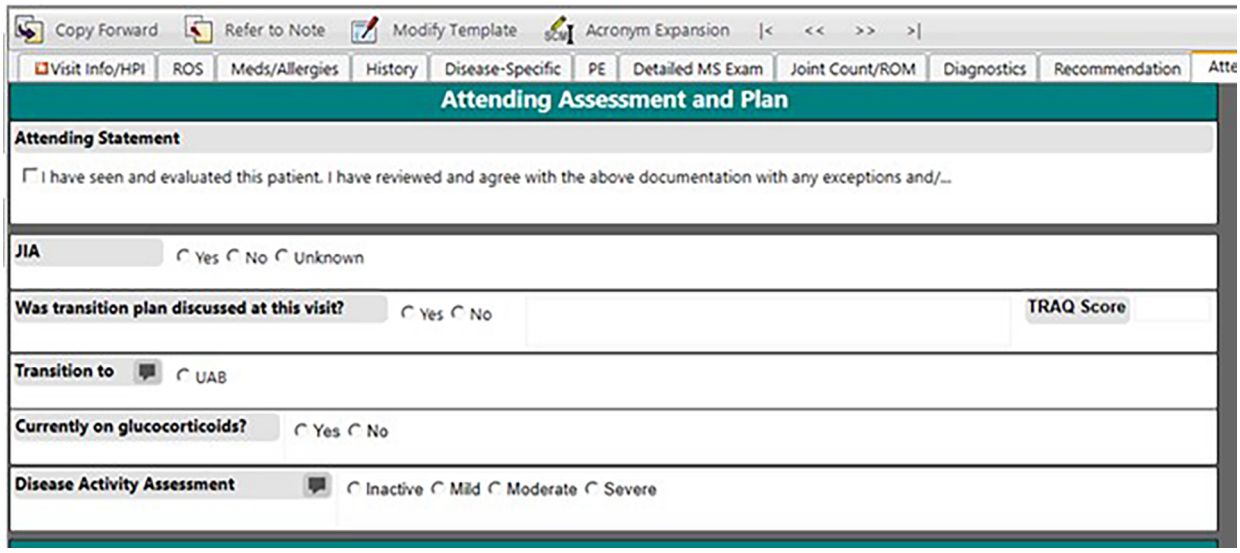

FIGURE 2
Screenshot of the EHR clinical note template with the provider-determined JIA field.

utilize the new structured field. Once we consistently had more than 90% accuracy for the "JIA checkbox", we abandoned comparing this list to the patient list based on ICD-10 codes.

Dashboard development

As IT's resources became overburdened, our weekly rheumatology query result delivery became inconsistent and the EHR did not allow for clinical personnel to perform data queries. Given our limited IT resources and the time constraints for manually extracting data, combined with the need for frequent and ongoing data updates to continuously improve patient care and quality improvement initiatives, we identified an alternative approach (Figure 1). In May 2020, the COA IT department created a secure, limited-access dashboard external to the EHR to provide immediate and real time access to the structured data that had previously been shared by weekly emailed data query reports (Figure 3). The dashboard was designed based on input from providers to prioritize critical data points such as cJADAS10 scores and medication usage. It provided the flexibility to add, drop, and modify metrics as needed based on our current monitoring initiatives and the most current literature and recommendations. Not all structured data elements were included on the dashboard as they all did not require population monitoring on a frequent basis. The column order of variables were determined by IT based upon the data extraction. However, the dashboard structure allowed us to export data to other programs in spreadsheet or comma separated variable format. This function allowed us to better sort, visualize, and share data using graphs and tables.

Initially the dashboard was populated by patients with JIA identified using ICD-10 codes, with similar results to the weekly data reports. With an expanding portfolio of quality improvement projects, we shifted the dashboard to include all patients seen in the rheumatology clinic and included the structured variable for provider determined JIA diagnosis. This adjustment allowed us to filter on "JIA yes" for continued monitoring of accuracy and completeness of patient identification and JIA-specific metrics. Inclusion of all patients seen in

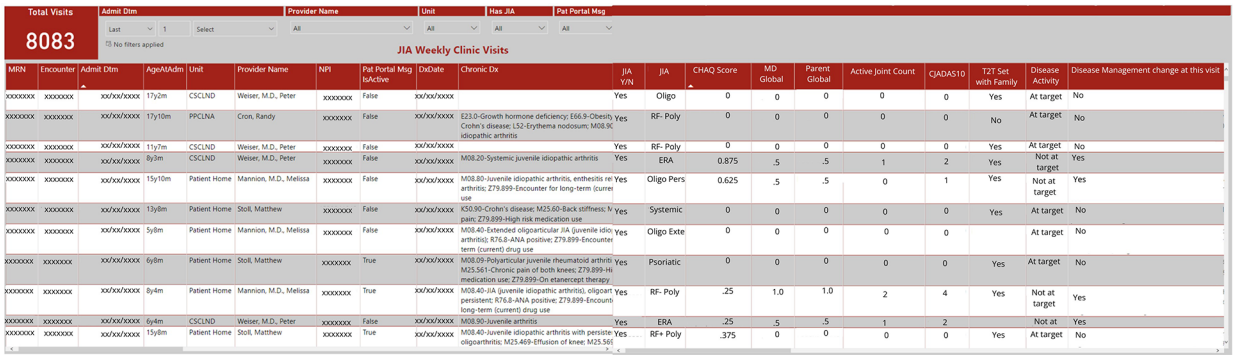

FIGURE 3
Screenshot of local dashboard populated by EHR data.

rheumatology clinic also allowed us to monitor other structured data from the clinic note which iteratively led to the modification and inclusion of additional structured data elements in the note.

Results

By modifying our outpatient rheumatology note in the EHR to include structured data elements and a JIA diagnosis attestation specific field, we were able to accurately identify our JIA patient population and collect disease activity measures while abandoning the use of manual chart abstraction. We identified an average of 309 patients per quarter, all subtypes were represented, and ages ranged from 12 months to 20 years. Encounters from our main clinic site as well as offsite and telemedicine visits were all included. Within 3 months of creation of the "JIA checkbox", we achieved and maintained 99% accuracy for identifying patients with JIA. These structured data elements also facilitated the development of an external dashboard based on the EHR that updates in real time to allow us to monitor documentation, track quality improvement initiatives, and eliminate the need for recurring data requests. By easily monitoring quality and disease measures in a specific population, we can track outcomes and processes that have been implemented into our clinic (Figure 3). The structured data elements are also automatically extracted to populate an external learning network registry to further our local and national improvement initiatives. COA/UAB pediatric rheumatology was the first and only site with a non-Epic EHR to successfully implement electronic data transfer to the PR-COIN Registry.

In addition to improving and maintaining reliable utilization of the "JIA checkbox" to identify an accurate JIA population, our team used the structured data elements and available dashboard for other improvement initiatives. For example, in conjunction with PR-COIN (21), we began to monitor use of treat to target (T2 T) processes for our patients with JIA. This included setting a treatment goal with the patient and family, calculating the cJADAS at the point of care, and assessing whether the patient was at goal or not at goal. During the course of the network-wide initiative, we were able to increase documentation of a T2T goal from 0 to 85% of visits for JIA and improved cJADAS documentation from 17 to 88% of visits. We accomplished this through weekly monitoring, multiple PDSA cycles, and division-wide celebrations for achieving our smart aim goals. Following this initiative, by improving our measurement of cJADAS and documenting a T2T goal, we had an overall reduction in the mean cJADAS across our JIA population, from 4.0 in April 2019 to 2.3 in January 2023. This reduction indicates an overall improvement in disease activity including patient outcomes, reflecting the effectiveness of measuring a disease activity score in achieving lower disease activity levels that is seen and recommended in other diseases like rheumatoid arthritis (22).

Discussion

The structured collection of data, frequent monitoring, and continuous improvement of quality metrics are crucial elements in the modern healthcare setting, especially in managing chronic conditions like JIA. This initiative demonstrates how advancements in EHR systems can significantly enhance the management and treatment outcomes of such diseases through efficient data utilization. We customized our rheumatology outpatient EHR note with structured data fields to populate a real-time dashboard that enabled us to improve documentation of quality metrics and improve disease activity measures in our JIA patient population. By utilizing the EHR to collect electronic clinical quality measures that have been tailored to our practice, automatic extraction allowed for more efficient generation of performance measures. The dashboard allows for frequent performance updates and development of targeted improvement strategies.

We demonstrated improvement in documentation of the components of the cJADAS and T2T goal setting for patients with JIA and saw an improvement in average cJADAS across our JIA population over time. This reduction indicates that patients are experiencing fewer symptoms and less severe disease activity, overall contributing to better disease control and improved quality of life for the population. While difficult to interpret the

significance of change for an individual in this population since there are patients with both oligo- and polyarticular disease, the reduction in score for the population is an important metric for quality care in the clinic (14). Improvement in the population cJADAS could be a result of improved disease activity or improved health related quality of life (23) through optimized treatment, increased response to adverse effects of medication, addition of non-pharmacologic treatments, increased education of the score itself, and awareness of monitoring by both patients and providers resulting in a social desirability bias. Frequent disease activity assessment is a component of the recommended T2T approach for the management of JIA to quickly achieve disease control and limit long-term complications of disease (18). Other components include target disease activity setting with the patient, and treatment changes to achieve the disease activity target (18). Disease activity measures can be used for each individual at the point of care in a T2T approach (21), but these measures can also be used to assess the disease activity of a clinic population in evaluation of overall quality of care. Importantly, by monitoring our documentation performance we were able to maintain high levels of T2T goal setting even after the primary intervention ended.

The introduction of a JIA-specific attestation field within the outpatient rheumatology notes has not only streamlined the process of patient identification but has also reduced the inaccuracies associated with the reliance on ICD-10 codes alone. Initial efforts to collect and monitor metrics were time consuming because of limited resources and resulted in a lack of consistent and accurate data. Because ICD-10 codes and administrative claims are not always the most accurate way of determining primary disease (24), we improved our process for identifying patients with JIA within our local EHR. Implementation of modified EHR systems has been shown to streamline the documentation process, standardize data entry procedures, and improve data accuracy and completeness. These modifications have been instrumental in addressing longstanding challenges associated with manual documentation, such as illegibility, inconsistency, and fragmentation of patient records (1, 4, 25). The addition of other structured data fields allowed for critical information such as disease activity, treatment responses, and patient well-being to be consistently recorded and easily accessible. This structured approach facilitates more accurate population health management and individual patient care, highlighting its significance in clinical settings (1, 26).

Dashboards are a tool used to extract data to make information easily accessible to the user. They originally were primarily utilized in the marketing field; however recently they have been modified to become a valuable resource in the healthcare field (4). Dashboards can present individual or population level information in a timely manner and can be flexible to allow inclusion of metrics that are important to the department, clinic, and patients. The information displayed can demonstrate change in one measure over time, change in measures in response to an intervention, or a cross sectional assessment of several measures at one time (4, 27–30). In this case we utilized the population-based real-time dashboard to identify interventions for rapid Plan-Do-Study-Act cycles and monitored documentation performance and subsequent changes in clinical outcomes for the cohort.

Moreover, the structured nature of EHR templates allows for standardized data capture, facilitating easier aggregation and analysis of clinical data for research and quality improvement purposes. As a result, healthcare organizations have been able to leverage EHR-derived data to monitor clinical performance, identify areas for improvement, and implement targeted interventions to enhance the quality and safety of patient care delivery (4, 29). Through the systematic collection of patient data, we were able to generate comprehensive datasets to monitor our documentation performance and evaluate patient outcomes over time to identify areas for improvement to enhance the quality and safety of patient care delivery. Data collected during routine clinical care not only benefits individual patients but also contributes to broader research and quality improvement efforts aimed at enhancing healthcare outcomes on a population level. We also included structured data fields critical for the PR-COIN Registry, initially for ease of manual data entry, but the structured format allowed for automated electronic data transfer.

Challenges to the development and continued utilization of the electronic dashboard include resources to be able to make necessary changes, continuation of processes, and limited data availability within the EHR. With the lack of resources available to address concerns or problems as they arise, we may wait weeks for problems to be resolved. Other limitations that exist include dependency of providers to continue to mark the disease specific attestation checkbox for patients. Although we achieved >90% accuracy and providers only need to mark yes once, new diagnoses will require providers to continue to participate in this process. We have continued to monitor accuracy monthly and remind providers as needed to sustain our performance. Additionally, our data collection is limited to the information that has been collected in the current EHR, introduced to our local outpatient clinics in 2017. Any data from prior to 2017 still requires manual chart abstraction and is subject to high rates of missing data.

The advancements in EHR technology and its application in our local pediatric rheumatology clinic exemplify the potential of digital health solutions to revolutionize medical care. Structured data collection ensures that relevant clinical information is accurately captured and organized, supporting comprehensive and personalized patient care. Frequent monitoring through innovative tools like dashboards enables real-time data analysis, essential for effective disease management and intervention adjustments. The focus on quality metric improvement helps in refining clinical practices and enhancing patient outcomes. We have continued to modify the outpatient note to create structured values that will transfer into the dashboard as needed to meet the needs of various QI projects and research studies. The ability to measure and evaluate performance in real-time will allow us to improve other quality metrics in other patient populations.

Data availability statement

The datasets presented in this article are not readily available because these data are directly linked to the EHR and are not

deidentified outside of the counts presented in the article. Requests to access the datasets should be directed to Melissa Mannion, mmannion@uabmc.edu.

Author contributions

LT: Conceptualization, Data curation, Investigation, Project administration, Writing – review & editing. HD: Data curation, Methodology, Project administration, Software, Writing – review & editing. NM: Data curation, Methodology, Project administration, Software, Writing – review & editing. ES: Conceptualization, Data curation, Investigation, Project administration, Writing – review & editing. MM: Conceptualization, Investigation, Methodology, Project administration, Supervision, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Improving lupus care index documentation in patients with childhood-onset systemic lupus erythematosus

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Introduction: Childhood-onset systemic lupus erythematosus (c-SLE) presents unique challenges due to increased risk for severe morbidity and mortality compared to adult-onset SLE. Effective disease management relies on accurate disease assessment and documentation. Our project aimed to improve the documentation of the Lupus Care Index (LCI), a disease assessment bundle, by implementing a quality improvement (QI) initiative.
Methods: A QI project was conducted at Nationwide Children's Hospital (NCH), targeting patients with c-SLE. The LCI, comprising the Systemic Lupus Erythematosus Disease Activity Index (SLEDAI-2k) Physician Global Assessment (PGA) and patient-reported pain score, was introduced to capture comprehensive disease assessment. Interventions included provider education, standardization of documentation procedures, and electronic health record (EHR) modifications. Automated reports tracked documentation rates, and Pareto charts identified areas for targeted interventions.
Results: Baseline analysis revealed incomplete documentation of LCI components in only one-third of c-SLE patients. Following interventions, documentation rates improved from 38% to 90%, with sustained improvement over at least a year.
Discussion: Enhancing documentation of LCI in patients with c-SLE is crucial for optimizing disease management. Our quality improvement initiative demonstrated the feasibility of improving documentation practices through targeted interventions and system modifications. Future research should explore the impact of comprehensive documentation on clinical outcomes in pediatric lupus patients. Improving documentation of LCI in patients with c-SLE is essential for optimizing care delivery and clinical outcomes; our QI initiative highlights the effectiveness of systemic interventions in enhancing documentation practices and underscores the importance of continued efforts to improve pediatric lupus care.

KEYWORDS

childhood lupus, SLE, systemic lupus erythematosus, SLEDAI-2K, PGA, quality improvement

Introduction

Systemic lupus erythematosus (SLE) is an autoimmune disease that affects multiple vital organs, such as the brain and the kidneys. SLE affects adults and children, with an estimated prevalence of 3.3–8.8/100,000 in children (1). SLE is associated with significant morbidity and mortality and a more aggressive disease course in Childhood-onset SLE (c-SLE) (2). There is an increased risk of early mortality among patients with childhood vs. adult-onset SLE (3), with the most common causes attributed to active disease and organ failure (4, 5). Adequate control of the disease reduces the risk of organ damage, which is more common in children (6). Reliable measurement and documentation of disease activity are key initial steps in disease control (7). Our team developed a Lupus assessment bundle called the Lupus Care Index (LCI) to capture lupus disease activity. This index utilizes three existing metrics: Systemic Lupus Erythematosus Disease Activity Index (SLEDAI-2K) (8), Physician Global Assessment of Disease Activity (PGA), and patient-reported pain score. At Nationwide Children's Hospital, we found that only one-third of patients with c-SLE had complete documentation of all three components of LCI. We implemented a quality improvement project to increase Lupus Care Index documentation from 38% to 80%, with sustained improvement for at least 1 year.

Methods

Context

Nationwide Children's Hospital (NCH) is a 470-bed, free-standing academic medical center in Columbus, Ohio. From 2016 to 2017, the NCH rheumatology clinic had 6,906 outpatient visits. About 100 childhood-onset SLE (c-SLE) patients are seen yearly in a multidisciplinary specialized lupus clinic and general rheumatology clinics. The multidisciplinary lupus clinic includes rheumatologists, nephrologists, pulmonologists, specialty pharmacists, psychologists, neuropsychologists, and a social worker. In addition, the rheumatology team comprises nurses, administrative staff, and quality improvement (QI) data specialists.

Intervention(s)

Our team developed a disease activity assessment bundle called Lupus Care Index (LCI) to provide overall disease assessment. LCI utilizes three existing metrics: Systemic Lupus Erythematosus Disease Activity Index (SLEDAI- 2K), Physician Global Assessment (PGA) of disease activity, and patient pain score [0–10 on Visual Analog Score (VAS) or Wong-Baker FACES^{TM}].). SLEDAI 2K is a weighted index in which signs and symptoms, laboratory tests, and physician's assessment for each of the nine organ systems are given a weighted score and added with a score range of 0–105, with higher scores representing greater disease activity. Items are scored if present during the visit or within 30 days before or after the visit (9). PGA is a physician's assessment of the severity of disease based on a 10-point Likert scale (score of 0 = no disease activity and 10 = very high disease activity). Rheumatologists may document SLEDAI-2K and PGA after the clinic visits. However, these are not completed consistently during every visit. Patients self-report their pain score, which reflects their average pain score related to lupus over the past week before the clinic visit. It is obtained based on FACES^{TM} or VAS and recorded by nurses upon patient intake and before starting clinic encounters.

Quality improvement (QI) team

A QI team was established, which included six pediatric rheumatologists, rheumatology fellows, a nurse and a nurse lead, a QI specialist staff member, a psychologist, and an administrative staff member. This QI team met monthly as part of QI team meetings, focusing on multiple projects related to childhood lupus.

Inclusion/exclusion criteria

The baseline analysis included patients with c-SLE receiving medical care in the pediatric rheumatology clinic at NCH from January 1, 2016, to November 31, 2016. We excluded clinic visits scheduled for teaching purposes, such as new medication injection teaching appointments.

Plan-do-study-act (PDSA) cycles

The QI team met in January 2018 to brainstorm ideas revolving around significant areas of improvement for documenting the bundle elements. A key driver diagram was developed to identify the major factors that impacted physicians' and nurses' documentation of LCI. The key drivers contributing to achieving our goals included raising provider awareness of the LCI to ensure that all healthcare providers were fully informed about the importance and methodology of documenting LCI scores. The second driver included standardized scoring by creating consistent and reliable processes for measuring and recording LCI using indices such as SLEDAI-2K and PGA. The third key driver included documentation practice improvement by implementing systems and tools to facilitate thorough and accurate recording of relevant patient information.

Our first PDSA cycle focused on establishing requirements for the LCI and educating providers by sharing clear guidelines and protocols for providers to follow, ensuring uniformity in documentation practices across the board, and on how and where they could provide documentation of disease activity indices in the electronic health record. Consensus was established among team members that SLEDAI- 2K should reflect clinical and laboratory data from the 30 days (10) before or after the clinic visit and PGA as assessed by the provider during the

intended visit. Nursing staff reviewed charts daily to ensure proper documentation of pain scores. Lastly, we developed an electronic health record (EHR) soft stop to remind physicians to document disease activity before closing their clinic charts. The QI team lead provided timely performance feedback through monthly reports, helping providers identify and address gaps in documentation.

Study of the intervention(s)

We ran monthly reports tracking SLEDAI completion and emailed physician providers and the nursing team with timely performance feedback. We provided reports using p-charts (Figures 1 and 2) showing the percentage of c-SLE patients who had complete documentation of all three components of LCI. In addition, we ran Pareto charts (Figure 3) to assist with a targeted intervention approach focusing on the least documented indices.

Measures

Patients with lupus whose clinic visit included documentation of all three components (SLEDAI-2k, PGA, pain score) of the LCI following their clinic visit were considered to have complete documentation. Documentation was considered incomplete if one or more elements were missing. The outcome of interest was the percentage of return lupus patient visits with complete documentation of LCI.

Analysis

LCI documentation was evaluated using a control chart. Pareto charts were utilized to identify deficiencies in the documentation of individual components.

Ethical aspects

Documentation of disease activity involves process improvement to improve the quality and safety of medical care. Therefore, the project was considered IRB-exempt.

Results

As the awareness of the project started in November-December 2017, we noted a slight increase in documentation during December. A slight non-significant improvement over time was noted after applying the first PDSA cycle. Root cause analysis helped us understand that initial reporting included patient clinic visits that were not closed, likely because some SLEDAI

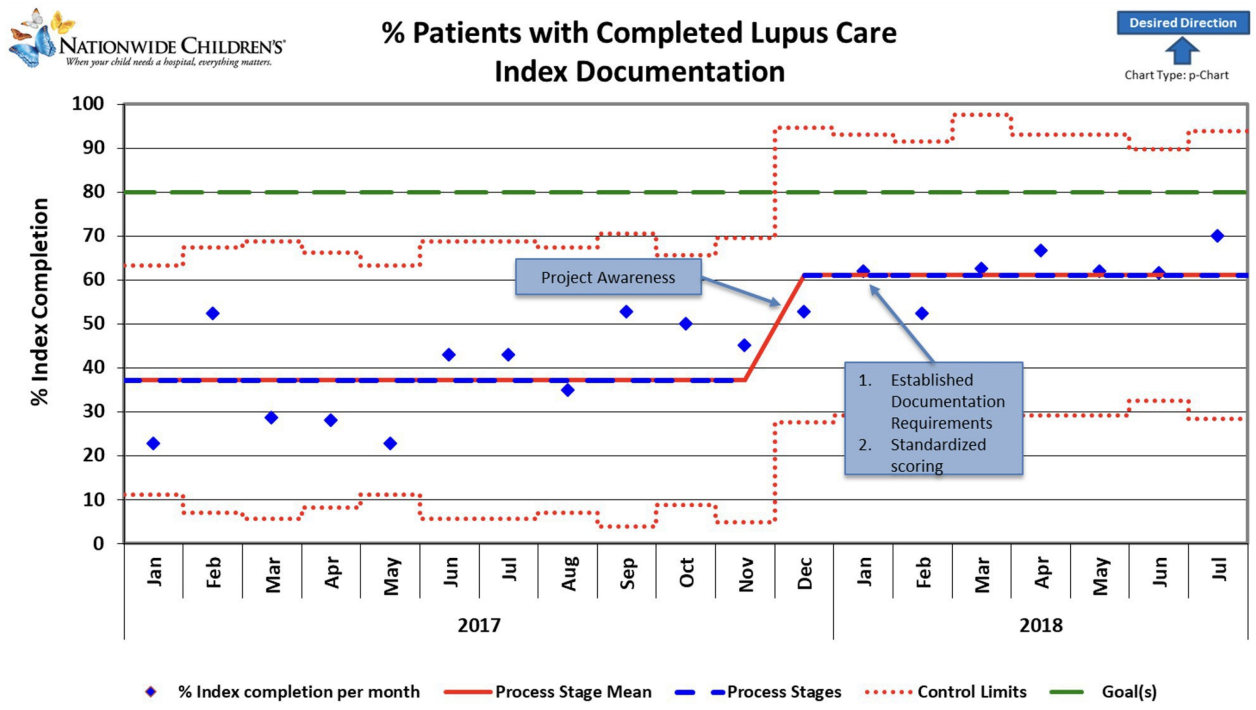

FIGURE 1
Control chart showing change over time during the project period. Blue diamonds represent individual data points. Red lines indicate control limits, while green lines mark a target performance level. The dashed red line fluctuates, indicating variable control limits. Notable milestones include increased project awareness, resulting in an upward trend and a sustained, stable performance. Improvements correlate with establishing standardized scoring and documentation requirements, contributing to consistent performance gains. The graph's general trend indicates a positive direction toward the desired outcomes, highlighted by the upward-pointing arrow.

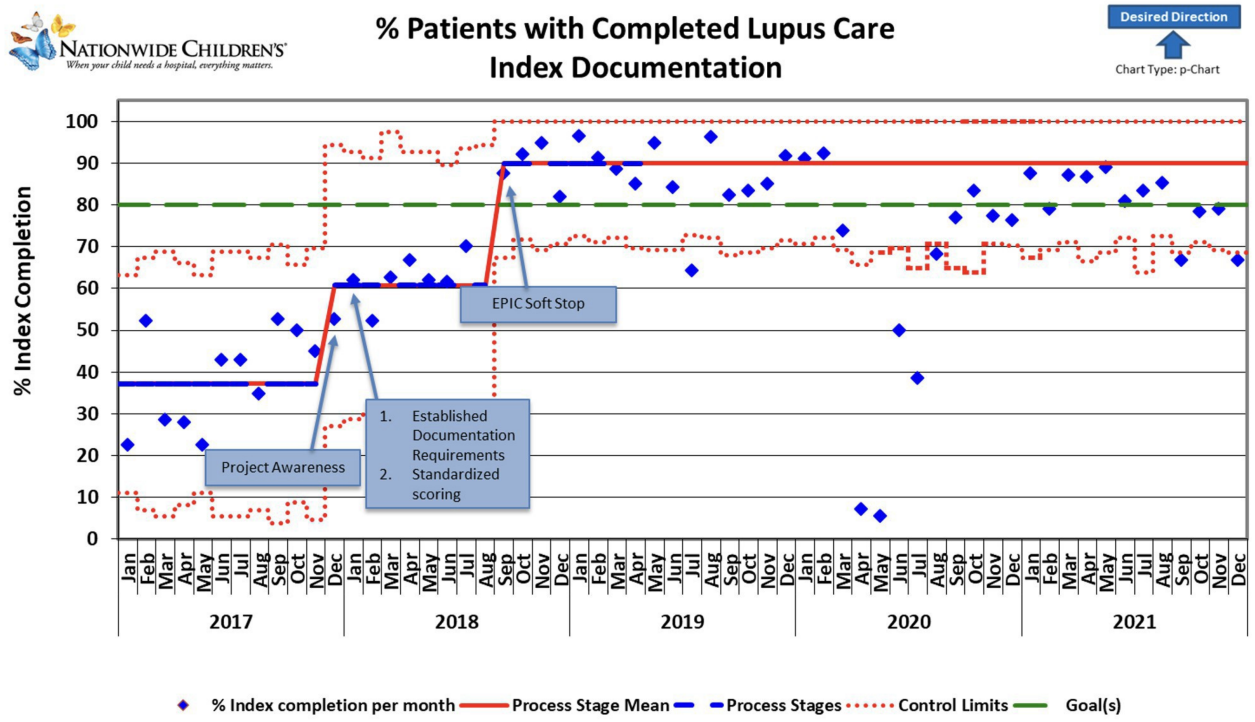

FIGURE 2
The control chart shows changes over time during the project period and extends beyond the project period to show sustained improvement. The reduction in documentation in 2020 was attributed to the SARS-CoV pandemic; due to expected ongoing pandemic impacts, recalculations of the Process Stage Mean line were not performed beyond March 2020. Blue diamonds represent individual data points. Red lines indicate control limits, while green lines mark the target performance level. The dashed red line fluctuates, indicating variable control limits. Notable milestones include increased project awareness, resulting in an upward trend and sustained, stable performance persistence of higher documentation rates beyond the project period. Improvements correlate with established standardized scoring and documentation requirements, contributing to consistent performance gains. The graph's general trend indicates a positive direction toward the desired outcome, highlighted by an upward-pointing arrow.

FIGURE 3
Comparison of document completion rates for lupus care index components. This before-and-after bar chart displays the percent baseline data (Nov-17) and individual component improvement Jun-18) across different parts of the LCI bundle.

laboratory components were unavailable for a week or more after the encounter ended. Thus, in August 2018, monthly reporting was changed to the middle of the following month to allow time for those results to be documented and charts closed. Our final PDSA was to make an EHR change to include a soft stop in September 2018 to alert providers about missing SLEDAI documentation. This PDSA resulted in an improvement of documentation from 61% to 90% (Figure 1). Our improvement has been sustained for over 1 year, as shown in Figure 2.

Discussion

In managing childhood-onset systemic lupus erythematosus (c-SLE), it is critical to recognize the heightened risk of end-organ damage and mortality compared to adult-onset disease. Thus, a primary objective is to minimize disease activity and prevent disease-related harm, aligning with a "treat to target" approach (11). Recent research underscores the feasibility and benefits of maintaining low disease activity states, as evidenced by improved clinical outcomes and prognosis.

This quality improvement (QI) initiative demonstrates the feasibility of improving the documentation of disease activity status in children with c-SLE at every visit utilizing the LCI bundle concept. The project aimed to improve the documentation rates of all three components of the LCI bundle from 38% to 80%. It has shown a promising trajectory with early indications of improved comprehensive charting practices. Utilization of the LCI is an innovative approach, integrating existing measures such as SLEDAI-2K, PGA, and patient pain scores to offer a comprehensive lupus disease assessment, which includes a patient-reported outcome, the Pain score. The initial modest rise in documentation following the implementation of the first PDSA cycle underlines the complexity of changing clinical documentation habits. However, the subsequent modification to our monthly reporting strategy and the introduction of an EHR soft stop in September 2018 has led to a more substantial and sustained improvement in practice.

One of the key insights from this project is recognizing the gap between the ideal and the practical in clinical documentation. The root cause analysis highlighted that the delay in laboratory results was a significant barrier to timely documentation. Addressing this barrier by adjusting the reporting period to include laboratory results suggests that flexible system design is crucial in achieving QI goals. Subsequent steps have implemented an automated report to capture laboratory results to reduce the provider burden of entering the results in the EHR after they are available.

Several limitations must be acknowledged. This QI project did not control all variables that could affect documentation practices, such as changes in clinic staffing or patient volume. Ongoing reporting has revealed gaps with incomplete documentation, missing laboratory values, and gaps in urine collection, which are being addressed through ongoing efforts. Additionally, the focus on documentation as a quality measure may not directly correlate with improved patient outcomes. Future research should explore the impact of comprehensive disease activity documentation on clinical outcomes in patients with c-SLE.

Our QI effort highlights the significance and complexities inherent in systematically documenting and managing disease activity in pediatric lupus care. By introducing the LCI alongside targeted educational and system-based interventions, we can enhance the quality of care delivered to pediatric lupus patients. Sustaining these efforts and conducting further studies are imperative to grasp these enhancements' impact on patient care comprehensively. It is important to note that this project has also improved data collection and utilization for c-SLE research, as evidenced by the recent initiative by the Childhood Arthritis and Rheumatology Research Alliance (CARRA) (12).

This QI project on documentation of LCI bundle assessing lupus disease activity status is the initial step. Moving forward, we plan to leverage this documentation to adopt validated criteria such as Lupus Low Disease Activity Status (LLDAS) and implement targeted interventions for managing c-SLE. Our Ultimate goal is to enhance the quality of life and optimize care delivery and clinical outcomes for children with c-SLE while mitigating the morbidity and mortality associated with this intricate autoimmune condition.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material, further inquiries can be directed to the corresponding author.

Author contributions

F-BS: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Supervision, Validation, Writing – original draft, Writing – review & editing. C-YT: Conceptualization, Data curation, Project administration, Writing – review & editing. SA: Data curation, Writing – review & editing, Project administration. OA: Conceptualization, Data curation, Writing – review & editing, Project administration. SA: Conceptualization, Methodology, Project administration, Writing – review & editing. AL: Data curation, Software, Writing – review & editing. EO: Conceptualization, Methodology, Writing – review & editing. VS: Conceptualization, Data curation, Methodology, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Automated mental health screening in pediatric lupus: associations with disease features and treatment

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Introduction: Patients with childhood-onset systemic lupus erythematosus (c-SLE) have higher rates of depression than their peers, which has been associated with worse medical outcomes. Therefore, it is imperative that their mental health be addressed. We utilized quality improvement (QI) methodology to automate mental health screening for patients with lupus within a pediatric rheumatology clinic. The retrospective cohort study aims to evaluate the association between mental health screening outcomes and demographics, medications, and disease activity measures in patients with childhood lupus.

Methods: The mental health QI team at a quaternary pediatric rheumatology center implemented an automated process for mental health screening in patients with c-SLE. Patients seen between 2017 and June 2023 with a diagnosis of c-SLE were identified using International Classification of Disease -Clinical Modification (ICD-CM) codes. Disease activity was assessed with the Systemic Lupus Erythematosus Disease Activity Index 2000 (SLEDAI 2K). Medications were identified on outpatient and inpatient orders for conventional synthetic and biologic disease-modifying anti-rheumatic drugs, hydroxychloroquine, corticosteroids, and aspirin. Mental health screening was accomplished with the Patient Health Questionnaire (PHQ). Descriptive statistics, univariate and multivariate linear regression were used.

Results: Between January 2017 and June 2023, 117 patients with c-SLE (41% with lupus nephritis) completed 534 total screenings. Each patient completed PHQ screenings, a median of 5 [interquartile range 2, 6] times. Screening increased when the screening process was automated. Those who were Black, female, or prescribed leflunomide, mycophenolate, and corticosteroids had higher PHQ scores.

Conclusions: Mental health support is essential for patients with chronic rheumatologic diseases such as SLE. Sustainable processes for quickly identifying depression are needed for optimal care of patients with SLE. Our process of automated, streamlined mental health screening successfully increased the screening of patients with SLE at every visit and led to timely interventions for positive PHQ scores. Higher PHQ scores were correlated with patients on leflunomide, mycophenolate, and corticosteroids. Future research should identify modifiable risk factors for high PHQ scores that the medical team can target.

KEYWORDS

lupus, pediatric, depression, mental health, screening, patient health questionnaire, patient-reported outcomes, informatics

1 Introduction

Addressing mental health is essential to optimally care for and treat patients with systemic lupus erythematosus (SLE). Pediatric and adult patients with SLE have higher rates of depression and anxiety than the general population (1–5). Children with SLE have 2.9 times increased odds of being diagnosed with depression and have 5.4 times increase in suicidal ideation (4). The reported prevalence of depression in childhood-onset SLE (c-SLE) is 20%–59% (5), compared to 24% in adults (6).

In addition to its prevalence, pediatric patients with more severe depression have increased lupus disease activity, cardiovascular disease, physical disability, suicidal ideation, premature mortality, and lower educational attainment (7). In general, patients with depression are three times more likely to have medication non-compliance than their non-depressed counterparts (8), and increased medication non-adherence is associated with worsening depression symptoms in patients with c-SLE (5).

Given these outcomes, a survey of members of the Childhood Arthritis and Rheumatology Research Alliance (CARRA) reported that 95% of responding pediatric rheumatologists supported mental health screening every 6–12 months. However, only 7% of this cohort routinely screened symptomatic patients, and only 2% screened all patients with a standardized, validated tool (9). While, in general, providers in pediatric rheumatology recognize the need for addressing mental health, implementation of screening is lacking. This discrepancy is likely due, in part, to inefficient screening methods and the fact that the current screening approaches may not be sufficient to address mental health needs in this population (10).

In this study, we describe the quality improvement efforts at a large quaternary children's hospital to automate and streamline mental health screening, making this essential and potentially time-consuming process feasible in a busy clinic setting. We also compared these mental health screening scores to patient demographics, immunosuppressive medications, and lupus disease activity.

2 Materials and methods

2.1 Setting

Nationwide Children's Hospital (NCH) is a large pediatric quaternary care academic medical center. The rheumatology team at NCH comprises pediatric rheumatologists, pediatric rheumatology fellows, a nurse practitioner, a social worker, nurses, a pharmacist, and a clinical psychologist. Our team utilized quality improvement methodology to develop an automated screening process to assess depression in patients with c-SLE (11, 12). We then retrospectively evaluated the data collected between January 2017 and June 2023. This study was approved by the Institutional Review Board (STUDY00003317).

Patient Population: Patients evaluated at a large quaternary care hospital outpatient rheumatology clinic with a diagnosis of c-SLE were identified using the respective International Classification of Disease—Clinical Modification (ICD-CM) codes 710 (ICD9) and M32 (ICD10).

2.2 Patient characteristics

Patient demographics were extracted from the electronic health record, which included sex, race, and ethnicity. Race was categorized as White, Asian, Black, Multiple, or Other/Unknown. Nephritis was defined as a patient with an ICD-9 or ICD-10 code for lupus nephritis. The specific ICD codes utilized were 710.0, 583.81, 710.0, 583.89, M32.14, and M32.15. Date of diagnosis and date of Patient Health Questionnaire (PHQ)-8 completion were recorded, and disease duration was calculated.

2.3 Disease activity

Systemic Lupus Erythematosus Disease Activity Index 2000 (SLEDAI 2 K) was calculated at each standard of care visit (13). In some instances, the SLEDAI 2 K was not fully completed when initially recorded due to pending lab values during clinical care and was later calculated via automated processes. For urinary values, if epithelial cells were present, the sample was considered contaminated, and hematuria and pyuria were not recorded unless this was documented as due to active disease by the provider. Similarly, if hematuria was present, manual chart review was completed to evaluate menstruation status. Patient and provider-reported disease activity assessments were recorded on a standardized 0–10 scale, with higher score indicating worse disease (14). These scores have been recommended by the American College of Rheumatology and the Outcome Measures in Rheumatology Consensus Initiative for use to more fully evaluate the overall health of patients with rheumatologic conditions (15, 16).

2.4 Medications

Outpatient and inpatient orders for conventional synthetic and biologic disease-modifying anti-rheumatic drugs, hydroxychloroquine, corticosteroids, and aspirin were identified; the first and last orders for each medication were identified, and consistent use was assumed. Cyclophosphamide exposure was defined as the first and last dates of consecutive infusions, with no more than 120 days between administered doses, plus 28 days from the last dose; this broad administration window would account for medication being held, such as in the case of an infection. Rituximab exposure was defined as the first and last dates of consecutive rituximab infusion occurring within 40 days of each administered dose plus six months from the last dose, as we assume rituximab would have an effect for approximately six months after the last dose. One patient could have multiple courses of cyclophosphamide and/or rituximab exposures.

2.5 Mental health screening with patient health questionnaire (PHQ)-8

Our team initially utilized the PHQ-9 for depression screening. The PHQ-9 includes a self-harm question; our questionnaire included additional self-harm questions including "Has there been a time in the past month when you had serious thoughts about ending your life?" and "Have you ever, in your whole life, tried to kill yourself or made a suicide attempt?". In 2023, the PHQ-8 and Ask Suicide Questionnaire replaced the PHQ-9 and additional self-harm questions, to be more complete when evaluating suicidality. Of those who completed the PHQ-9, their scores were recalculated to only capture PHQ-8 questions. The PHQ-8 will hereafter be referred to as PHQ. Due to the critical need to act on high scores or an indication of suicidality, a PHQ was not given to the patient until he or she physically arrives at clinic. PHQ scores of 0–4, 5–9, 10–14, and 15–19, and 20 or greater indicate none, mild, moderate, moderately severe, and severe depression, respectively (17). Previous quality improvement interventions included increasing rheumatology providers' awareness of screening by discussing it at staff meetings, streamlining the workflow of mental health screening for social work and the nursing team, integration of identifying patients to be screened into nursing pre-visit planning, and increasing patient and family awareness of the screening project and mental health issues in rheumatology. These interventions resulted in annual, routine PHQ screening in 2017 on paper for all English-speaking patients with lupus ≥12 years old; responses were transcribed into the electronic health record (11). In 2021, our final QI cycle automated the screening process. The PHQ questionnaire was transitioned from a paper form to being delivered electronically on a tablet to all English-speaking patients with lupus ≥12 years of age at every visit; this questionnaire was automatically assigned to the clinic encounter and only available upon checking into clinic.

After full integration into the electronic health record (EHR), PHQ scores were automatically calculated and populated in the clinic note with a drop-down menu of options indicating the action taken. Patients with a PHQ score of 5–9 were provided a handout focusing on psychoeducation and contact information for the rheumatology psychosocial team, including social worker and psychologist. A PHQ score of ten or higher would trigger an intrusive pop-up alert in the EHR when providers, including physicians, nurse practitioners, social workers, and psychologists, open the patient's chart. The alert would then be acknowledged, and the provider would address the concern or contact a social worker or psychologist if they were unaware. A social worker or rheumatology psychologist would then meet with the patient during the clinic visit. A thorough suicide risk assessment would

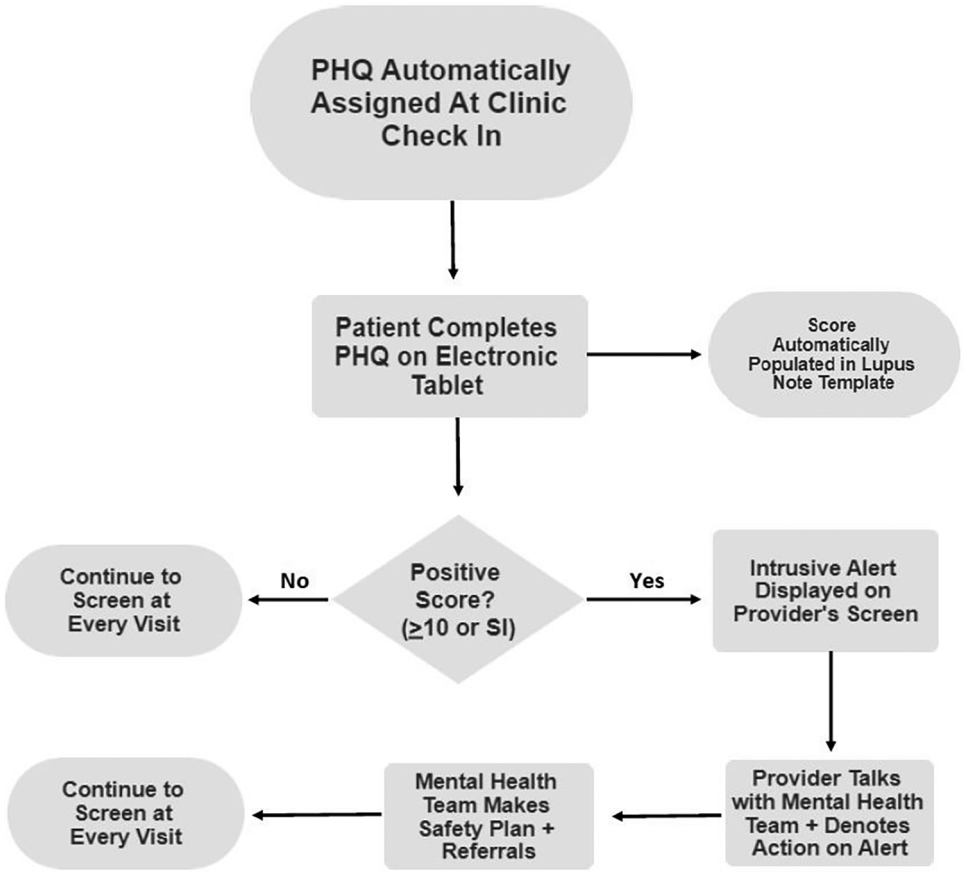

FIGURE 1
Clinical workflow for critical alerts. PHQ, Patient Health Questionnaire; SI, suicidal ideation.

be done with the Ask Suicide Questionnaire and Columbia Suicide Severity Rating Scale, and the patient would be offered a behavioral health referral (Figure 1) (18, 19).

2.6 Provider feedback

Five months after complete automation, providers were asked "How satisfied are you with the current automated depression screening for patients with lupus" on a 0–10 ordinal scale, with anchors of "0-Not Satisfied" and "10-Very Satisfied".

2.7 Statistical analysis

Summary statistics describing the population are reported as median [interquartile range (IQR)] and count (percent). Univariate regression evaluated the association between PHQ scores, patient, disease characteristics, activity scores, and treatments. Variables with $p < 0.20$ in univariate were evaluated in multivariate linear regression modeling. Analysis was completed using Stata 16.0.

3 Results

One hundred seventeen unique patients (41% with lupus nephritis) completed 534 screenings (Table 1). Each patient completed PHQ screenings a median of 5 [IQR: 2, 6] times. Screening frequency increased after electronic implementation. A mean of 50 screens were completed annually between 2017 and 2021; screens increased to 191 in 2022 and the first two quarters of 2023, with a median score of 2 [0, 7]. Figure 2 depicts median PHQ scores by year. Those with a new diagnosis within the prior 6 months had an average PHQ score of 2 [0,6] whereas those diagnosed more than 6 months ago had an average PHQ score of 3 [0, 7]. There was no statistical significance between the two groups ($p = 0.45$). Of the patients who completed a PHQ, eight (4%) reported a suicide attempt, and two (1%) had suicidal thoughts within the past month. PHQ scores were 347 (64%), 99 (19%), 64 (12%), 20 (4%), and 4 (1%) indicating none, mild, moderate, moderately severe, and severe scores, respectively. After complete automation, intrusive alerts fired appropriately for PHQ scores 10 or higher. This alert fired 84 times at 23 visits. It occurred from 1 to 9 times per visit, as it would be triggered each time a provider newly entered the chart. A provider acted on the alert at every visit. During all but three visits, either a social worker or psychologist met with the patient to follow up on the elevated PHQ score. In these three visits without follow-up, the patients were already connected with psychology, psychiatry, or counseling, and none were actively suicidal. Of the 12 providers surveyed, the median satisfaction of the new automated screening was 10 [9, 10].

In univariate analysis, leflunomide, hydroxychloroquine, mycophenolate, corticosteroids, female sex, Asian and Black race, Hispanic ethnicity, patient global, provider global, and SLEDAI 2K were associated with higher PHQ scores (Table 2). In multivariate analysis, Black race and patient global scores were associated with higher PHQ scores provider (Table 3).

TABLE 1 Demographics, disease activity, and mental health screening by presence of nephritis.

	All patients ($N = 534$)	Without nephritis ($N = 316$)	With nephritis ($N = 218$)	*P*-value
PHQ Score by Year				0.21
2017	2 [1, 7]	4 [1, 7]	2 [1, 5]	
2018	4 [2, 10]	4 [1, 10]	4 [2, 10]	
2019	4 [1, 7]	2 [0, 7]	5 [3, 9]	
2020	5 [2, 10]	4 [1, 8]	7 [3, 10]	
2021	3 [1, 9]	3 [1, 10]	2 [0, 7]	
2022	2 [0, 6]	0 [0, 5]	3 [0, 7]	
2023	1 [1, 4]	0 [0, 4]	3 [0, 7]	
Age	17 [16, 19]	18 [16, 19]	17 [15, 19]	<0.01
Female sex	434 (81%)	257 (81%)	177 (81%)	0.98
Race				0.02
Asian	57 (11%)	33 (11%)	24 (11%)	
Black	193 (36%)	121 (38%)	72 (33%)	
Multiple	33 (6%)	18 (6%)	15 (7%)	
Other/ Unknown	34 (6%)	12 (3%)	22 (10%)	
White	217 (41%)	132 (42%)	85 (39%)	
Hispanic ethnicity	49 (9%)	23 (7%)	26 (11%)	0.07
Patient global[a]	1 [0, 4]	1 [0, 4]	1 [0, 3]	0.53
Provider global[b]	1 [0, 2]	1 [0, 1]	1 [0, 3]	<0.01
SLEDAI 2K[c]	2 [0, 6]	2 [0, 4]	4 [0, 8]	<0.01

PHQ, patient health questionnaire; SLEDAI 2K, systemic lupus erythematosus disease activity score 2000.
[a]Available in 145 encounters.
[b]Available in 318 encounters.
[c]Available in 374 encounters.

4 Discussion

Our study shows that depression is highly prevalent in those with c-SLE, as more than one-third of PHQ screenings were at least 10 or higher, which indicates a likelihood ratio of 7.1 and specificity of 88% for depression per a PHQ validity study (17). We sought to streamline the mental health screening process to better detect depression, thereby optimizing care for children with SLE. Automating PHQ screening and embedding the process in the electronic medical record ensured consistent screening and that every positive screen was addressed, reducing the burden on clinic staff. This process solves many of the frequently cited obstacles in the CARRA mental health survey: time constraints and increased burden on staff (9). Our embedded collaborative psychology and social work teams within the rheumatology clinic were instrumental to complete these screenings as they allowed us to act upon positive screens without the need for transfer to the emergency room or behavioral health to obtain additional evaluation and safety plan initiation. We

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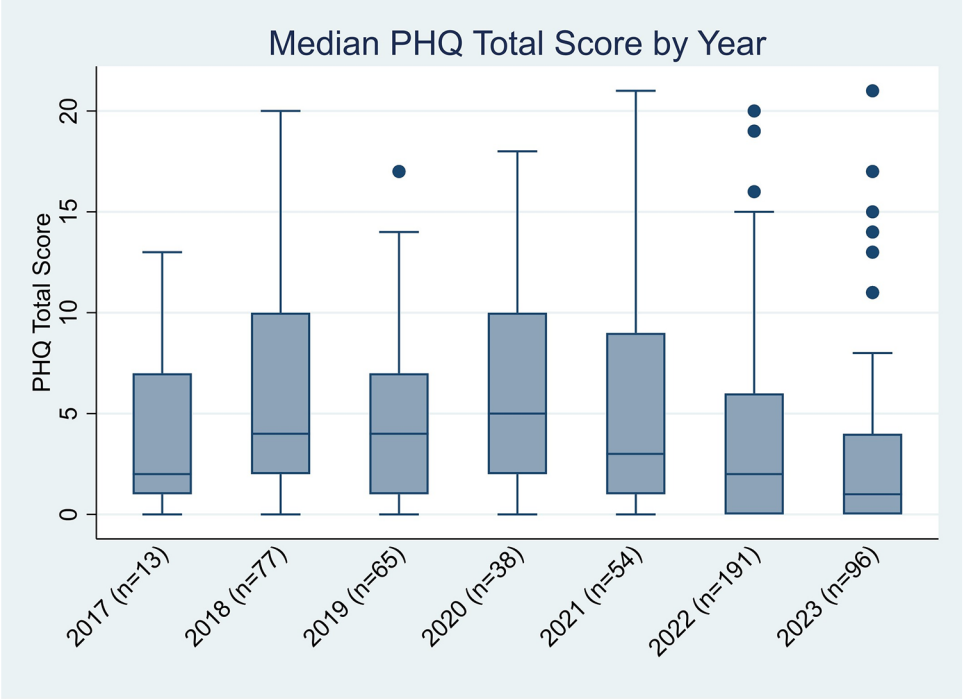

FIGURE 2
Median PHQ scores by year. PHQ, Patient Health Questionnaire; SLE, systemic lupus erythematosus.

TABLE 2 Univariate linear regression of Patient Health Questionnaire 8 by medication use, demographics, and disease activity score.

	β-coefficient (95% CI)	*P*-value
Hydroxychloroquine	0.95 (0.08, 1.81)	0.03
Sulfasalazine	1.82 (−1.58, 5.24)	0.29
Leflunomide	13.83 (1.31, 23.34)	<0.01
Methotrexate	−1.00 (−2.47, 0.47)	0.18
Mycophenolate	1.92 (1.01, 2.82)	<0.01
Azathioprine	0.17 (−0.87, 1.21)	0.75
Tacrolimus	3.07 (−1.72, 7.87)	0.21
Intravenous immunoglobulin	2.81 (−3.97, 9.59)	0.42
Corticosteroids	1.31 (0.49, 2.14)	<0.01
Abatacept	1.85 (−0.84, 4.53)	0.18
Aspirin	−0.47 (−2.64, 1.72)	0.68
Belimumab	1.20 (−0.42, 2.84)	0.15
Rituximab	0.89 (−0.85, 2.63)	0.32
Cyclophosphamide	1.06 (−0.74, 2.85)	0.25
Age	0.04 (−0.12, 0.20)	0.60
Female sex	1.47 (0.42, 2.53)	<0.01
Race		
White	Reference	Reference
Asian	−1.70 (−3.06, −0.33)	0.02
Black	2.40 (1.49, 3.31)	<0.01
Multiple	0.19 (−1.53, 1.91)	0.83
Other/Unknown	−0.60 (−2.29, 1.10)	0.49
Hispanic ethnicity	1.54 (0.12, 2.97)	0.03
Patient global	0.77 (0.60, 1.15)	<0.01
Provider global	0.55 (0.21, 0.90)	<0.01
SLEDAI 2K	0.17 (0.07, 0.26)	<0.01

CI, confidence intervals; SLEDAI 2K, systemic lupus erythematosus disease activity score 2000.

successfully increased screening from an annual screen to screening at every visit with the implementation of automated questionnaires. One of the biggest challenges we faced was the COVID-19 pandemic and the need for telehealth visits and the screening frequency decreased.

TABLE 3 Multivariate linear regression of Patient Health Questionnaire by medication use, demographics, and disease activity score.[a]

	β-coefficient (95% CI)	*P*-value
Hydroxychloroquine	0.91 (−0.58, 2.39)	0.23
Leflunomide	—	—
Methotrexate	0.69 (−1.35, 2.75)	0.50
Mycophenolate	1.21 (−0.22, 2.65)	0.10
Corticosteroids	−0.22 (−1.66, 1.21)	0.76
Abatacept	−6.27 (−11.49, 1.04)	0.02
Belimumab	−1.19 (−3.74, 1.36)	0.35
Female sex	−0.34 (−1.86, 1.17)	0.66
Race		
White	Reference	Reference
Asian	1.63 (−0.22, 3.47)	0.08
Black	2.28 (0.77, 3.79)	<0.01
Multiple	1.82 (−1.87, 5.51)	0.33
Other/Unknown	4.52 (0.80, 8.23)	0.02
Hispanic ethnicity	1.59 (−1.77, 4.96)	0.35
Patient global	0.81 (0.52, 1.09)	<0.01
Provider global	0.52 (0.08, 0.96)	0.02
SLEDAI 2K	−0.07 (−0.20, 0.06)	0.20

CI, confidence intervals; SLEDAI 2K, systemic lupus erythematosus disease activity score 2000.

[a]Model reduced to 132 encounters.

This study also evaluated relationships between mental health screening results, demographics, medications, and lupus disease activity measures. Our study showed that leflunomide, mycophenolate, and corticosteroids, was associated with higher PHQ scores, i.e., worse mental health. Steroid use is associated with mood dysregulation and depression (20), and patients requiring steroids typically use this medication early in the disease course or during flares. Therefore, steroids are needed during more stressful times when patients' disease is active, and they do not feel well. However, we did not see similar associations with cyclophosphamide or rituximab, commonly used during active disease. Interestingly, higher PHQ scores and belimumab use were not significantly correlated despite documented concerns that belimumab may worsen depression and suicidality (21). Our results support those of a meta-analysis of randomized control trials of belimumab use for patients with SLE, which did not find that depression or suicidality increased in patients taking belimumab (22). Another contributing factor may be that prescribers avoid starting belimumab for patients with known depression, who would be at higher risk for elevated PHQ scores.

We also found that lower patient global and provider global scores were associated with lower PHQ scores. This finding is not surprising, given that lower scores indicate less active disease, and when patients are less symptomatic and feel better, they may have lower rates of depression. This is in alignment with previous studies showing that those in remission have less depression, less anxiety, and improved health-related quality of life (23, 24).

Patients who were Black were significantly more likely to have high PHQ scores ($p < 0.01$). Although this relationship between depression and increased PHQ scores has been previously noted in minority populations, minorities are less likely to receive counseling or other support services for their mental health than non-Latino White children (25). This finding likely has multifactorial implications involving social determinants of health and inequities in the healthcare system. For example, another study found that being a person of color, attaining a lower level of education, being unmarried, not having medical insurance, and being unemployed were all associated with higher PHQ scores. Interestingly, when accounting for other socioeconomic factors, Black race was associated with higher PHQ scores, which highlights the complexity of evaluating social constructs (26). Given this intricate interplay, social determinants of health must be considered when addressing mental health. In addition, more efforts must address disparities in mental health care between White populations and people of color.

Few patients reported a suicide attempt and suicidal thoughts, 8 (4%) and 2 (1%), respectively. Consistent screening would identify these patients so interventions could be quickly implemented. While these results are serious and necessitate action, this small number also highlights that, based on our results, we would not expect an overwhelming number of critical screens in a pediatric rheumatology office.

This study was limited by its setting, i.e., a single-center initiative in a large children's hospital with a social worker and psychologist embedded in the rheumatology clinic. Similar screening can be performed without these resources using automated screening with provider alerts for critical results. However, timely access to mental health resources will be vital to implementing a similar process in other institutions. The PHQ assesses symptoms of depression and suicidality, but it does not diagnose major depressive disorder per the Diagnostic and Statistical Manual of Mental Disorders (27). However, the PHQ is commonly used to screen for mental health concerns and is a validated screening tool (17).

Future research should assess whether modifiable risk factors can be identified for patients with SLE and depression. Analysis of social determinants of health specific to mental health scores may be informative. This analysis would ideally allow the medical team to intervene on those risk factors before patients develop significant depression or suicidality. In summary, it is clear that mental health support is essential for patients with chronic rheumatologic diseases such as SLE. Sustainable processes for timely identification of depression are needed to best take care of patients with SLE. Our process of automated, streamlined mental health screening successfully led to an increase in screening patients with lupus at every visit and in providing timely interventions for positive PHQ scores. Higher PHQ scores were correlated with patients being on leflunomide, mycophenolate, and corticosteroids. Future research should seek to identify modifiable risk factors for high PHQ scores that can be targeted by the medical team and to develop streamlined pathways for intervention.

5 Conclusions

Mental health support is essential for patients with chronic rheumatologic diseases such as SLE. Sustainable processes for quickly identifying depression are needed to best take care of patients with SLE. Our process of automated, streamlined mental health screening successfully increased screenings of patients with lupus at every visit and provided timely interventions for positive PHQ scores. Higher PHQ scores were correlated with patients on leflunomide, mycophenolate, and corticosteroids. Future research should identify modifiable risk factors for high PHQ scores so the medical team can target and develop streamlined intervention pathways.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material, further inquiries can be directed to the corresponding authors.

Ethics statement

The studies involving humans were approved by Institutional Review Board (STUDY00003317). The studies were conducted in accordance with the local legislation and institutional requirements.

The human samples used in this study were acquired from a by-product of routine care or industry. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin in accordance with the national legislation and institutional requirements.

Author contributions

LH: Writing – review & editing, Writing – original draft, Methodology, Conceptualization. SA: Writing – review & editing. AL: Writing – review & editing, Conceptualization. KD: Writing – review & editing, Conceptualization. VS: Writing – review & editing, Conceptualization. AT: Writing – review & editing, Writing – original draft, Supervision, Methodology, Formal Analysis, Conceptualization.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Navigating the path to equitable rheumatologic care for underserved children with quality improvement

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Objective: The aim of this quality improvement project is to identify children with rheumatologic conditions to prevent delayed or missed diagnosis in underserved pediatric populations. Our focus is on prompt and accurate identification and subsequent treatment of rheumatologic symptoms in pediatric patients referred from Atrium Health safety-net primary care clinics that deliver care to families without private insurance, including those lacking insurance entirely.
Methods: We collaborated with providers at one safety-net clinic to improve the processes of identification and subspecialty referral, resulting in an increase in the number of identified pediatric patients and referrals for these patients with potential rheumatologic disease. We used the Model for Improvement framework with rapid Plan–Do–Study–Act cycles and evaluated improvement with run and statistical process control charts.
Results: We achieved improvement, with zero referrals in the previous 5 years for the targeted population increasing to 15 patient referrals within 1 year of project initiation. Despite this increase in referrals, the rheumatology clinic was able to see all priority patients within 20 business days from referral.
Conclusion: An awareness of concerning rheumatologic symptoms in safety-net primary care clinics, combined with the use of both visual and decision aids, allows care teams to efficiently recognize and accurately refer patients needing specialty care.

KEYWORDS

health equity, specialty care, quality improvement, referral accuracy, care continuum

1 Introduction

Rheumatologic conditions are notoriously difficult to diagnose because of myriad symptoms affecting multiple organ systems, evolving over time before a definitive diagnosis is reached (1). Given their limited resources arising from financial problems and logistical barriers, health literacy burden, and demands from other types of family crises, patients from lower socioeconomic backgrounds have historically been subjected to the challenges and resulting consequences of delayed diagnosis and access to subspecialist care (2–5).

Founded in 2011, the Atrium Health Levine Children's Specialty Center Rheumatology (LCSC-RC) division serves both clinic outpatients and hospital inpatients. In 2020, Atrium Health started focusing attention on rectifying care inequities. LCSC-RC recognized this as an opportunity to use a quality improvement (QI) methodology to improve patient care. This project builds upon a prior QI project (6) that expedited referrals based on symptoms, with the current project further exploring and improving access for children with fewer resources available to them.

2 Methods

2.1 Aim and measures

The aim of this project was to identify underserved children with rheumatologic conditions to prevent delayed treatment or missed diagnosis. Our focus was on prompt and accurate identification and subsequent treatment of rheumatologic symptoms in pediatric patients referred from four Atrium Health safety-net primary care clinics (PCCs). The proxy measures that were taken are listed as follows:

- A 10% increase in referrals needing rheumatologist evaluation for pediatric patients from four Atrium Health safety-net clinics to LCSC-RC from September 2020 to September 2021.
- A total of 80% of all referred LCSC-RC patients scheduled to be seen within 30 business days from referral.

We used the Model for Improvement with rapid Plan–Do–Study–Act (PDSA) cycles as a framework. At project initiation, the core team, comprised of a pediatric rheumatology physician lead, resident lead, QI coach, and data analyst, secured partial grant funding. Given the project's aim to improve the quality of care locally, the Institutional Review Board approved the project as a Quality Improvement Project.

2.2 Problem description

To better understand the gap in pediatric rheumatologic care for those with limited resources, we reviewed data extracted from the electronic medical record (EMR) for pediatric patients who met all of the following criteria: (1) pediatric patients who received primary care services at Atrium Health between October 2015 and May 2020; (2) those living in either of two zip codes in the city designated as "public health priorities" due to higher rates of chronic diseases, infectious diseases, and deaths related to these conditions (7, 8); and (3) those presenting with symptoms indicative of rheumatologic disease. The third criterion was established previously to identify the group of symptoms most commonly found in referred patients, which ultimately led to a diagnosis of a rheumatologic condition, or at least required ongoing rheumatologic care (Figure 1, see priorities 1, 2, and 3) (6). The relevant symptom categories were joint pain, unexplained fever lasting >10 days, and positive antinuclear antibody (ANA). Four safety-net clinics emerged as seeing a large majority of patients from this search. These sites became our target population. Safety-net clinics deliver care to families without private insurance, including those lacking insurance entirely.

2.3 Baseline data

The review of our baseline data spanning 5 years revealed that of 2,239 qualifying patients, 1,929 (86%) were seen in four Atrium Health safety-net clinics and only five children (0.2%) ultimately consulted with a pediatric rheumatologist in our system. It was unknown whether the remainder of the qualifying patients were seen by a pediatric rheumatologist outside Atrium or whether the symptoms ultimately resolved without treatment.

As the COVID-19 pandemic erupted placing strict limitations on in-person interactions and severe burden on the healthcare system, the team pivoted from targeting all four safety-net clinics to only the main pediatric safety-net PCC where 858 of the 2,239 eligible patients were seen yet zero referred.

The PCC was also the best option due to its locational advantages: it was juxtaposed one block away from the core team's campus and it served as the ambulatory primary care rotation location for pediatric residents at Atrium Health's Carolinas Medical Center (CMC) where LCSC-RC was housed. All CMC pediatric residents work at the PCC on a weekly basis. The team recruited the PCC advanced practice provider (APP) to serve as a local champion.

Accordingly, the measure of 10% increase in referrals for pediatric patients needing rheumatologist evaluation from four Atrium Health safety-net clinics to LCSC-RC by September 2021 had been modified to a 10% (17 patients) increase in accurate referrals from the PCC by December 2021. This number was determined by dividing 858 patients (baseline) by 5 years, equaling 171 patients in 1 year; therefore, 17 patients is a 10% increase.

Likewise, and due to the loss of one pediatric rheumatologist, the metric of 80% of all referred LCSC-RC patients seen within 30 business days from referral had been modified to 80% of all priority patients seen within 30 business days.

Priority population is defined as patients requiring ongoing rheumatology care, that is, pediatric patients <18 years of age referred from another provider and requiring the expertise of a pediatric rheumatologist for care of a perceived rheumatologic/autoimmune condition with symptoms to include any of the following criteria: ANA titer >1:320, specific joint swelling or pain, persistent fever, or rash. Priority patients were further subclassified into priorities 1, 2, and 3 (see Figure 1).

Our main tools were a referral tool, several visual aids including key information, and timely progress reports with the teams. LCSC-RC created a templated information referral tool in the EMR for primary care providers to complete, designed a triage tool for the receiving rheumatology team to use, and redesigned visit categories to reserve high acuity appointments in a previous project focused on increasing overall access to rheumatology care (6). The referral algorithm offers decision support to help referring providers understand referral requirements and provide

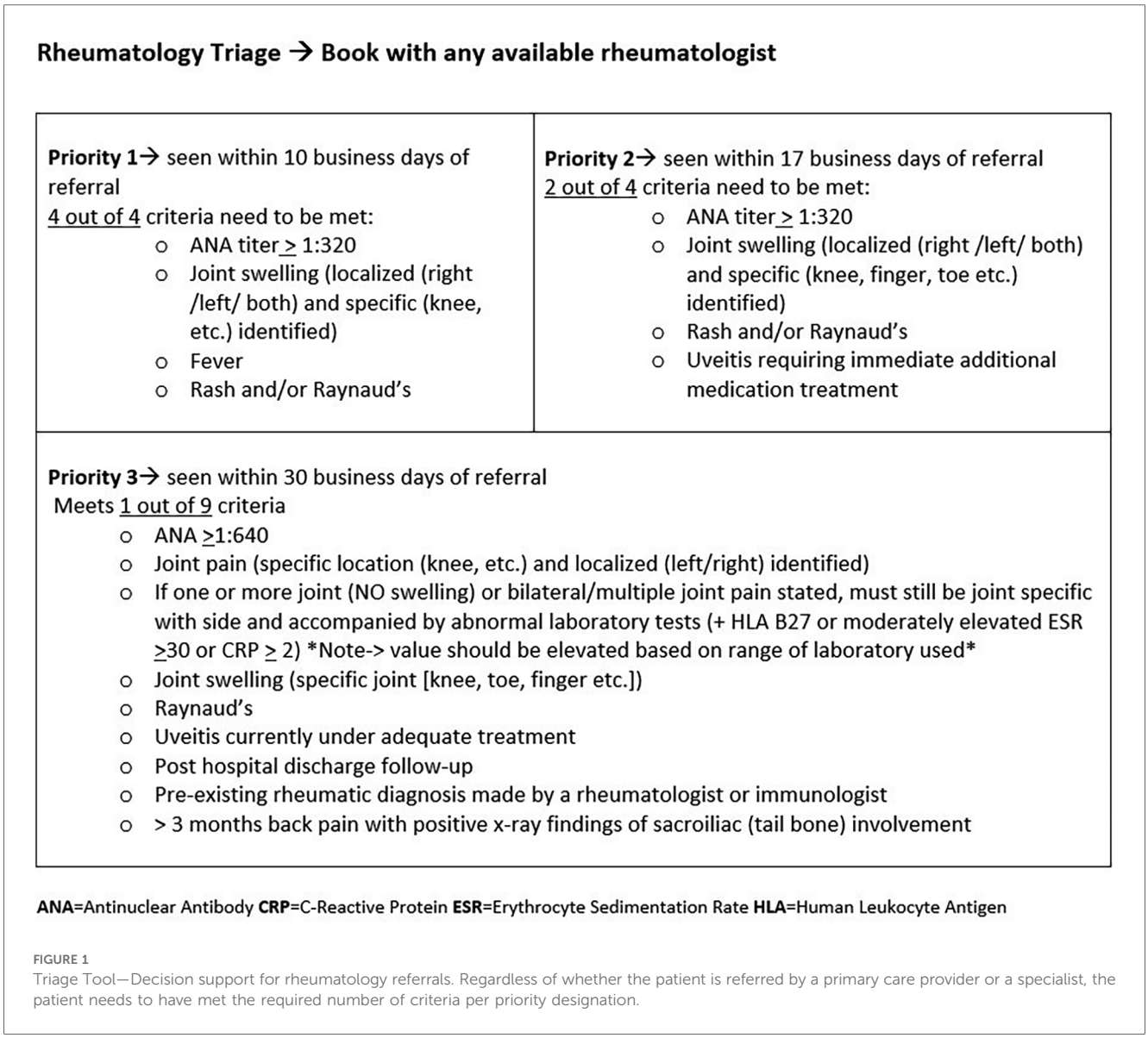

FIGURE 1
Triage Tool—Decision support for rheumatology referrals. Regardless of whether the patient is referred by a primary care provider or a specialist, the patient needs to have met the required number of criteria per priority designation.

specific information. The receiving referral coordinators then use the information to appropriately triage and schedule patients according to acuity. Incomplete forms trigger contact with the referring office and often include education. This allows for additional clarification of the requirements and ensures that patients are seen in a timely manner.

2.4 Change ideas

Change ideas included a focus on gaining primary care buy-in, team-to-team contact, training, and QI coaching. The team conducted multiple in-person visits at the PCC to develop a shared understanding of one another's work, to educate, and to test our tools. In October 2020, the LCSC-RC physician lead and the PCC APP met with PCC providers to introduce the project. The PCC providers requested decision support tools to facilitate appropriate identification of referral candidates. Returning the following month, the physician lead and APP introduced the PCC providers to the referral tool with a case study. PCC feedback endorsed the use of the referral tool combined with the case study and identified problems with removing outdated ICD 10 chronic diagnosis codes, their impact on workflow, and differences between attending physician use and resident physician use.

The resident developed visual aids to be displayed in three areas—on the wall, on computer keyboards, and on computer monitors—and in November 2020, the resident and APP determined that provider workrooms would be the most impactful locations. Flyers posted on the walls included triage priorities and instructions for following the referral pathway. Keyboard and monitor signs were small flags intended to reinforce behavior and provide EMR navigation support. The tools were placed in close proximity to one another. In December 2020, the physician lead shadowed the PCC providers

to better understand their workflow and ensured that the visual aids were visible and in appropriate locations.

In January 2021, the physician lead attempted to reconcile the ICD 10 chronic diagnosis codes, while the resident reintroduced the provider referral tool to her peers in February 2021. Based on feedback from the PCC providers, the project's resident and physician leads discovered that all providers needed more training to understand rheumatologic conditions and more durable, attention-grabbing visual aids. In March 2021, the visual aid posted on the workstation wall delineating referral-trigger symptoms and providing instructions for tool use was revised to incorporate bold fonts to highlight important items and different colors of background shading to separate symptom categories and was moved from behind computer monitors to a more visible space (Figure 2). The reminder taped to provider keyboards added pink font and was reinforced with stronger tape after it was discovered that the increasingly frequent sanitizing required during COVID made the original signs fall off quickly. Additional reminders were attached to the monitors.

The following months were dedicated to refreshing resident knowledge via their social media platform, and a division-wide resident-led grand rounds focused on the provider referral tool in April 2021. In the same month, the team turned their attention to previsit planning to better serve patients with language barriers. The triage tool of the rheumatology team was adjusted to capture language preferences, allowing an interpreter to be available for the appointment. Changes to the referral process for transportation needs and language barriers were embedded in a new EMR platform implemented in April 2022.

One innovation that required numerous PDSA cycles was the development of a culturally relevant patient story video paired with a referral tool tutorial video. The LCSC-RC physician lead had a teenaged patient from a Hispanic family who experienced a delayed diagnosis including multiple trips to the emergency room before being referred to LCSC-RC, resulting in a severe skin graft and hospitalization. The patient's story was particularly impactful, resulting in her serving as an advisor to the QI team. The team recorded this patient telling her story multiple times with intermittent testing at the PCC and with residents, improving scripting, focus, and audio/visual quality each time. The final product was combined with a previously produced instructional video of the physician lead explaining step by step how to access the referral tool through the EMR. This was shared electronically with the pediatrics leadership, office managers, and referral managers for the clinics involved and, through support from Atrium Health's marketing and communications office, with general pediatricians. These PDSA cycles spanned September 2021 through December 2021, with a special PDSA in September in which the resident shared the video with her resident peers and then conducted a survey on provider referral tool use to gauge knowledge retention, offering a gift card to incentivize survey completion. The provider lead also shared the video at a PCC monthly provider meeting in September.

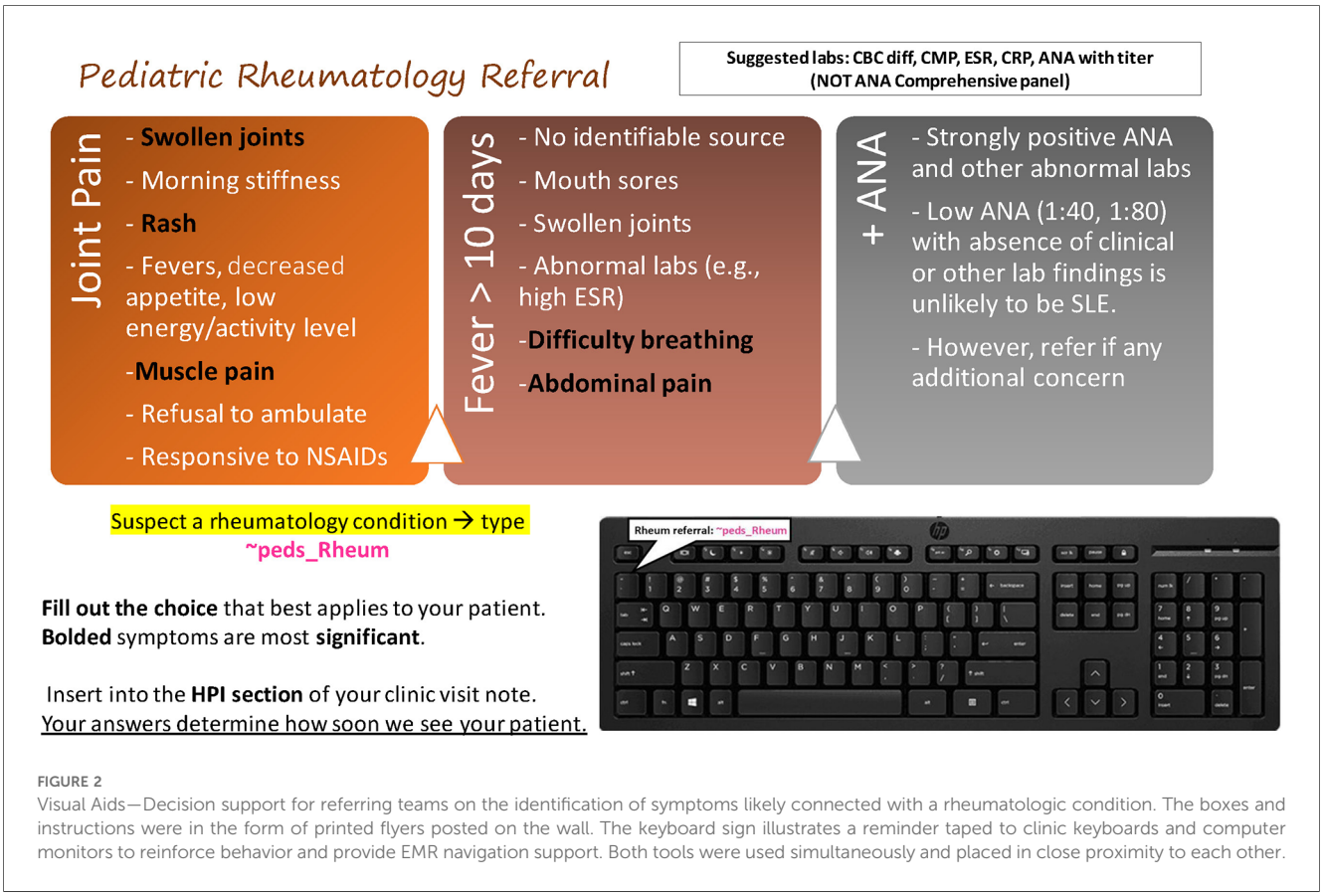

FIGURE 2
Visual Aids—Decision support for referring teams on the identification of symptoms likely connected with a rheumatologic condition. The boxes and instructions were in the form of printed flyers posted on the wall. The keyboard sign illustrates a reminder taped to clinic keyboards and computer monitors to reinforce behavior and provide EMR navigation support. Both tools were used simultaneously and placed in close proximity to each other.

3 Results

As mentioned previously, the team had to modify the intended target population and narrow it down to only the PCC. The first measure of 10% increase in referrals needing rheumatologist evaluation for pediatric patients fell short by two patients, receiving 15 of 17 targeted referrals. Eight out of the 15 referrals completed the referral tool. We still considered this very significant since the PCC had not referred any patients from the targeted population in the 5 years prior to the project (Figure 3). Significant interventions that helped to achieve non-random variation included the provider lead visiting the PCC to shadow clinic workflow and address specific referring provider questions (December 2020), updating the visual tool (March 2021), and sharing the patient story video to residents and referring providers (September 2021).

The second measure of 80% of all priority patients scheduled to be seen within 30 business days was achieved with an average of 20 business days from referral to consult (Figure 4). No special cause was observed. Other than the first patient referred, who was seen at 40 days from referral to consult, the goal of seeing referred patients within 30 days was surpassed and sustained at or above target for the remainder of the project.

Of the eight patients deemed priority, one was classified as priority 1, one as priority 2, and six as priority 3. Six were of Hispanic race and ethnicity, one was an African American, and one was an Asian, with the latter two having a non-Hispanic ethnicity. All eight patients had North Carolina Medicaid insurance. Their ages ranged from 7 to 16 years old, with four being males and four females. The final diagnoses given after a pediatric rheumatologist consult were hypermobility syndrome/hypermobile joints (5), pes planus (1), and systemic sclerosis (1). The 8th remaining patient received a scheduled appointment for a referral complaint of right-hand pain and ANA titer of 1:640 but then canceled their appointment, thus a diagnosis could not be made. The process itself was successful.

4 Discussion

This project found new ways to apply existing data analytics by layering symptoms data onto zip code data to uncover significant gaps in care. The team applied communication strategies to improve provider education and patient care, such as combining a culturally relevant patient story with a tutorial on referral tool use; using a resident and advanced practice provider to influence their peers; and helping all members of the care team collaborate effectively to maximize limited personnel and resources. Due to improved communication, provider knowledge, previsit planning, and process efficiency, the number of patients seen from disadvantaged communities increased despite COVID-19 limitations and severe staff shortages.

The visual aids acted as critical tools. Safety-net clinics face continuity disruptions, experience very high volumes, and often

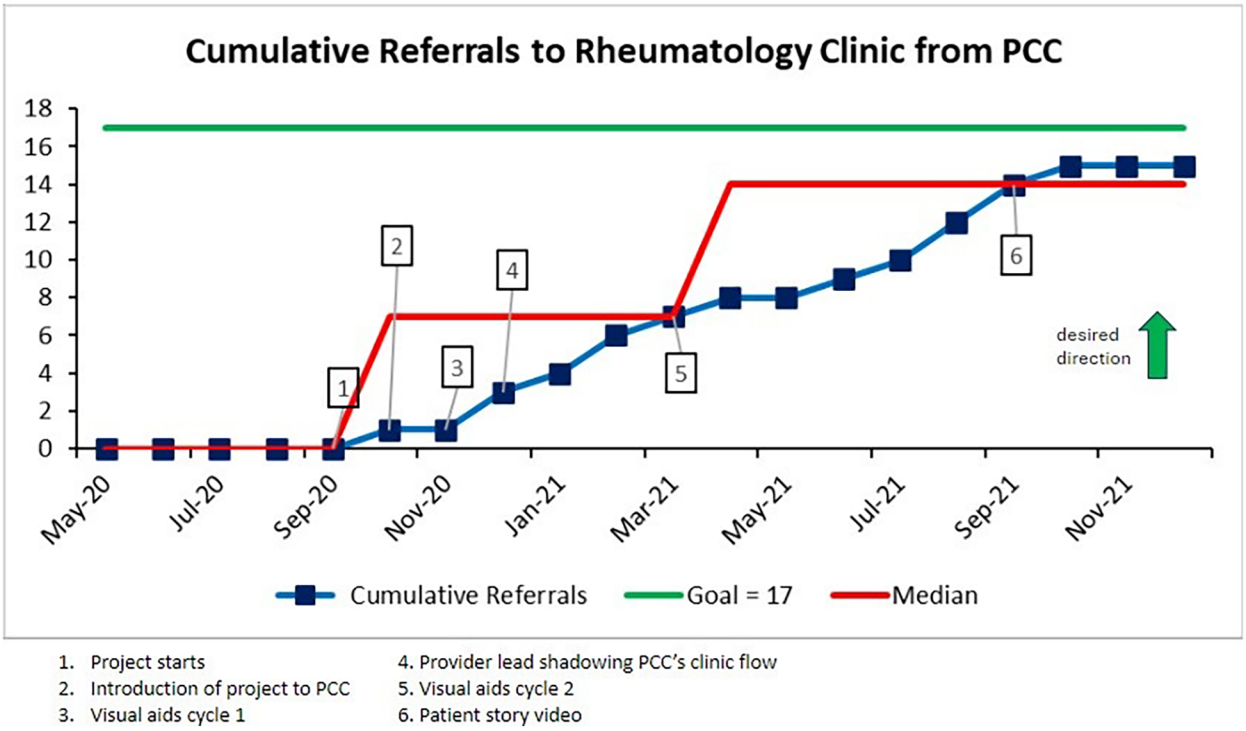

FIGURE 3
A run chart demonstrating cumulative referrals received from the target population. The referrals were evaluated on a monthly basis. The main interventions were noted. Two median shifts were observed based on the statistical rule of non-random variation of six data points above or below the median, ultimately reaching 15 total referrals.

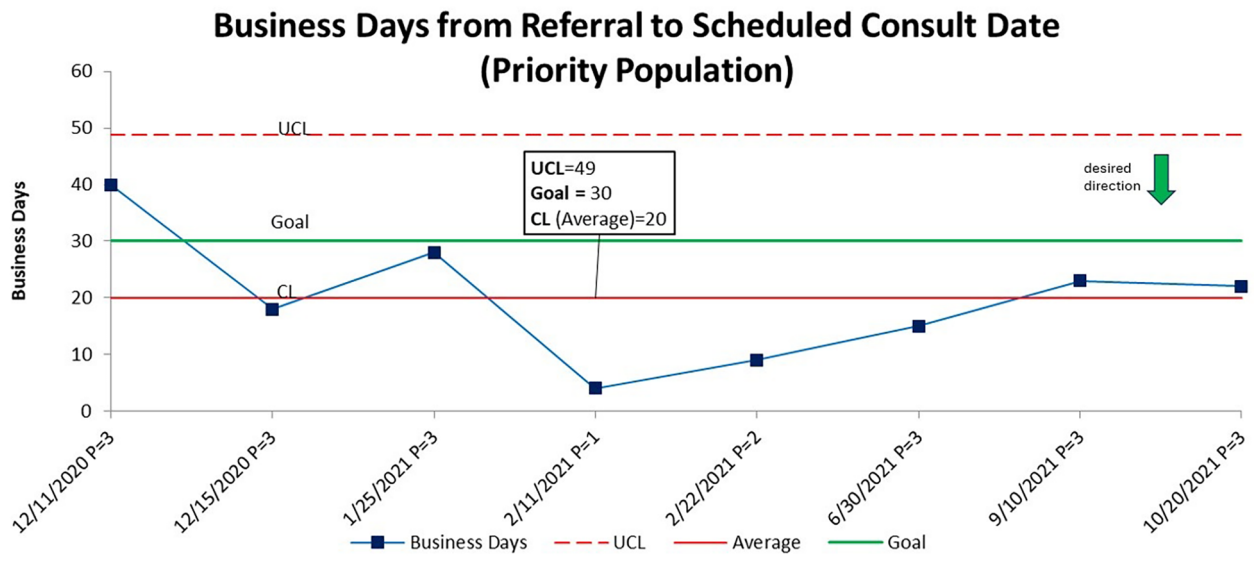

FIGURE 4
A statistical process control chart (SPC)—X chart demonstrating business days from referral to scheduled consult for priority populations. The center line (average) is stable at 20 business days. Each data point represents a patient and their level of priority. Only one patient missed the goal to be seen within 30 business days from referral to consult.

have rotating resident learners providing care, making diagnosis or identification of specific symptoms more challenging. Team visibility and direct contact helped partners recognize symptoms and take appropriate action. Most primary care providers had limited exposure to pediatric rheumatology during training (9), and the Atrium community is often aware of LCSC-RC only if they have a patient with a previous rheumatologic disease or interact with the team.

Including the referral coordinator team was extremely important, as this group ensured the closure of the referral loop. They had a vested interest in making the process flow smoothly, because problems disrupted their own work. The PCC referral coordinators were highly responsive, ensured that the PCC providers completed the referral tool, and met with the LCSC-RC lead physician twice virtually and once in person.

The resident lead and PCC APP were instrumental in project success, serving as the eyes and ears of project implementation at the PCC. This would be the case any time, due to their consistent frontline work in the environment undergoing improvement, but they became irreplaceable when COVID restrictions were in place. The high volume of patients in a safety-net clinic can also make it difficult to test interventions and change processes because of the time constraints of the staff. We needed to be very strategic when building our relationships with key stakeholders in the practice and connecting with individuals who could help influence culture change and build goodwill, remove barriers, and facilitate tests of change and the adoption of successful strategies.

Resident feedback indicated that using a patient's story highlighting the challenges of a delayed diagnosis provided a meaningful connect to purpose, reminding everyone of the importance of this project.

Language and transportation access developed into an important component of the care package thanks to this work. Previously, in-person interpreters were often unavailable if they had not been prebooked, and online options often resulted in clinical disruption because of poor or dropped internet connections and subpar information delivery. The previsit planning involved in this project ensured that in-person interpreters were available during appointments, and the heightened demand for this service justified additional staffing for the language services division. Likewise, during the course of this work, the team recognized that some of the patient families encountered clinic attendance barriers due to transportation. As a result, grant funding from the rheumatology QI community via PR-COIN (Pediatric Rheumatology—Care & Outcome Improvement Network) now supports these needs through solutions such as gas cards and ride share vouchers. Transportation and language barrier questions are now a standard part of the referral triage process, with appropriate solutions provided as needed.

The COVID-19 pandemic presented a variety of challenges. With frontline staff already overwhelmed by pandemic patient needs, staff shortages impacted both clinics because of COVID infection and redeployment. Restricted clinic access made it more difficult to learn about the PCC's workflow and establish the best way to seamlessly incorporate the referral tool into their processes.

COVID highlighted the care discrepancies that marginalized populations endured. For example, many Atrium Health clinics adopted social distancing and telemedicine protocols to protect both staff and patients, such as completing paperwork online, collecting patient history via video, and having patients wait in their personal vehicles. Many of these strategies were problematic for PCC families who had limited technology access, who had

language barriers, or who depended on public transportation. These restrictions can also impede the ability of a provider to understand the complicated nuances of rheumatologic symptoms.

Our attempt to reconcile ICD 10 chronic patient diagnosis codes in the EMR was a failed PDSA. While the best practice in the PCC is to regularly update the ICD 10 chronic patient diagnosis codes, we found that many PCC providers were not doing so consistently. The code list helps clarify potential diagnosis by illustrating patterns indicating chronic conditions. However, it proved extremely difficult for us to get the majority of providers to adopt the practice of updating the codes. Addressing this issue was beyond the scope of this project.

The importance of the primary care provider's understanding of symptom significance and referral process cannot be overstated for patients with social disparities, especially those with Medicaid insurance (a state-issued public insurance, typically provided to those who do not have or cannot afford private insurance). If a patient receives insurance from North Carolina Medicaid, they must receive a referral from a primary care provider before being able to see a specialist.

4.1 Limitations

A limitation in our project was the lack of completeness in approximately half of the referring providers' submission of referral questions. As a result of limited information in seven referrals, the rheumatology team triaged patients with pain in multiple joints at a higher sensitivity rate. Ultimately, this led to a diagnosis of hypermobility syndrome/hypermobile joints, which was our most commonly identified diagnosis; while this syndrome is not a strictly rheumatologic condition, these patients benefited from a rheumatologic evaluation and may require ongoing subspecialist management (6). Another limitation of this project was that we identified only those patients whose homes were located in two public health priority zip codes and were referred by a safety-net clinic. The Mecklenburg County Public Health Department identifies four additional public health priority zip codes; patients residing in these areas could be included in future work (6). In addition, our data extraction exercise did not capture individuals who may suffer from social disparities but who do not live in zip codes specifically identified as "marginalized."

4.2 Future directions

Our future plans include spreading this work to the other three safety-net clinics that were originally identified. All pediatric rheumatology clinics in our system are currently engaged in improving the accuracy of information relating to patient race and ethnicity in the EMR and ensuring its completeness, which is necessary to provide culturally appropriate resources and equitable access to tailored care. We received grant funding to conduct a primary qualitative research study using interviews and surveys with families from safety-net clinics to explore their experiences and barriers to care. Patients will provide insight into improving processes, contribute ideas for a patient-centered referral process, and participate in developing health literate printed materials and/or scripting. In the future, we plan to create additional videos of patients' journeys. We would like to work with patients to develop a health "passport" to facilitate navigation of their own health, the healthcare system, and complex conditions. This passport will be especially helpful for patients who see providers from multiple healthcare systems with different electronic medical record platforms. This passport will become a printed document that they will carry when returning multiple times to a provider for a variety of symptoms or entering a new area within the system.

Sustainability of this work was rendered challenging due to the collaborating safety-net PCC undergoing leadership change, nursing staff turnover, EMR transition, change in the referral process, and referral coordinator roles being redefined. The project required a re-evaluation to find a more sustainable path forward, and is currently undergoing a refresh. Specific activities include new resident involvement and pediatric leadership support to design innovative tools. Ongoing work in this renewed environment includes patient interviews, new visual tools, new EMR tools (smart phrase and ambulatory referral order), and regrouping with other community-wide health systems that have also acquired new EMR systems.

This project and its predecessor (6) illustrate that by leveraging clinical information systems to enhance gap identification to improve access to rheumatologic care, teams can continually drive and refine next steps and create a constant feedback loop to increase focus and knowledge in safety-net clinics for vulnerable populations. The lack of an efficient process and delayed diagnosis exacerbate disparities and impact outcomes, quality of life, patient/staff satisfaction, affordability, and resource allocation. With pediatric rheumatologists in such short supply, this work is crucial to ensuring that those children most in need receive timely access to subspecialist care. Navigating the future of this discipline requires flexibility to adapt and a willingness to embrace change, both of which benefit from and contribute to a learning health system. Sharing the strategies and lessons learned from this project can improve outcomes across the pediatric primary/subspecialty care spectrum.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material, and further inquiries can be directed to the corresponding author.

Ethics statement

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent was obtained from the individual(s), and minor(s)' legal guardian/next of kin, for the publication of any potentially identifiable images or data included in this article.

Author contributions

SV: Writing – review & editing, Writing – original draft. SM: Writing – review & editing, Writing – original draft. TB-M: Writing – review & editing, Writing – original draft.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Implementation of an automated transition readiness assessment in a pediatric rheumatology clinic

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Background: Failure of successful transition to adult care for adolescents and young adults with chronic rheumatic diseases negatively impacts their health and wellbeing. Transition of care is a vital and complex process within pediatric rheumatology that can be difficult to execute. Use of quality improvement (QI) and clinical informatics (CI) can help implement transition programs.
Local problem: Despite efforts to improve transition of care within our pediatric rheumatology clinic, it has been difficult to implement and sustain good transition practices including assessment of transition readiness. Using QI methodology and CI, this study aimed to improve transition readiness assessment from 12 to 30% and sustain for one year by surveying transitioning patients yearly.
Methods: A transition-focused QI team utilized methods endorsed by the Institute for Healthcare Improvement and leveraged CI to improve survey completion. Control charts of survey completion rates were tracked monthly. Descriptive statistics were used to analyze survey responses.
Interventions: Interventions focused on automation of patient surveys at regularly scheduled clinic visits.
Results: 1,265 questionnaires were administered to 1,158 distinct patients. Survey completion rose from a baseline of 12% to greater than 90% and was sustained over 18 months. Identified educational needs included health insurance, scheduling appointments, obtaining care outside of rheumatology clinic business hours, Electronic Health Record messaging, and refilling medications.
Conclusions: By leveraging CI and QI methodology, we were able to assess transition readiness in more than 90% of our patients and identify gaps in self-management. Process automation can create sustainable transition practices.

KEYWORDS

pediatric rheumatology, transition of care, transition readiness, quality improvement, clinical informatics, adolescents and young adults, pediatric rheumatic disease

Introduction

Transition from pediatric to adult rheumatology is a necessary process for many of our patients given that rheumatic diseases are often life-long chronic illnesses. Active disease and adverse outcomes around the time of transfer are present in a significant percentage of patients (1, 2) and some of these patients ultimately do not end up under the care of adult

rheumatologists. Successful transfer of care rates can be as low as 50% (3, 4). Patients who do not transition successfully and those with certain rheumatic diseases are at increased risk of mortality and poor health outcomes (1, 5). The transition process is complex and many pediatric rheumatology clinics struggle to effectively prepare adolescents and young adults (AYA) (6). Recognition of the need for strategic transition planning has been established for the past few decades and multiple societies including the American Academy of Pediatrics (AAP), the American Academy of Family Physicians (AAFP), the American College of Physicians (ACP), the European League Against Rheumatism (EULAR) and the Pediatric Rheumatology European Society (PRES) have guidelines surrounding transition of care (7, 8). Many of these guidelines utilize the GotTransition^{TM}approach which focuses on six core elements to transition (7). These six core elements include: development and dissemination of a transition policy; tracking and monitoring of transitioning patients; recurrent assessment of transition readiness skills and education to advance these skills; development of a healthcare transition plan with appropriate documentation for individual patients; transfer of care of a patient to adult practice; and finally, confirmation of transfer of care with an opportunity to solicit feedback from the patient. These six elements of transition have also been favorably viewed by AYA with rheumatic diseases. In focus groups, AYA reviewing these six elements reacted favorably to them and emphasized the importance of focusing on autonomy and independence to empower patients to advocate for themselves (9).

Transition of care has been *a priori*ty at our center for many years, but due to a busy and constantly evolving health system and team, it has been challenging to implement and sustain progress (10, 11). Finding ways to seamlessly integrate effective processes into practice is imperative. Assessing patient readiness to transition via a survey is a crucial step toward providing education on the skills needed for successful transition. These modifiable behaviors are deemed important for successful transition (12). The results of the transition readiness surveys are intended to be used to focus on education and self-management skills empowering our patients to gain autonomy in the healthcare system.

Using methods endorsed by the Institute for Healthcare Improvement (IHI) model for improvement (13), a multidisciplinary team within the Division of Rheumatology sought to improve transition by focusing on the six core elements for transition (3). Utilizing a systematic approach, different elements were targeted at various times. After successfully implementing the transition policy, the team shifted focus to address transition readiness. In January 2021, the transition quality improvement (QI) team created a new project aim with the goal of increasing yearly transition readiness assessment from 12 to 30% by December 2021 and sustaining for one year.

Methods

Context

Nationwide Children's Hospital is a large, quaternary care children's hospital in Columbus, Ohio. The Division of Pediatric Rheumatology sees more than 1,600 unique patients annually. It is staffed by 8 pediatric rheumatologists, 1 adult and pediatric rheumatologist, 1 nurse practitioner (NP), 4 pediatric rheumatology fellow physicians, 1 psychologist, 1 clinical pharmacist, 1 social worker, and 7–10 part-time rheumatology nurses. One of the rheumatologists is a dual trained physician informaticist who joined the group in 2021. A transition QI team has been in place for more than 15 years. The team includes representatives from the rheumatology providers and allied health professionals listed above. As recommended by GotTransition^{TM} (7), we defined patients aged 14 or older and seen at least 3 times in the rheumatology clinic as the target population of transitioning teens with rheumatic disease. A written transition policy was created in 2017. AYA receive a paper copy of the transition policy during the rooming process once they have reached the target population. Transition Readiness Assessment Questionnaires (14) were in use since 2011, but their usage was not consistent. In 2018, the QI team created a simplified transition readiness assessment in alignment with a hospital-wide initiative. The simplified questionnaire was given to the target population yearly, which we defined as every 12 months. This frequency was chosen to reduce survey burden and allow opportunity for responsive education at multiple time points. The transition readiness assessment consisting of 12 questions deemed critical by our transition QI team (Figure 1) was utilized. The questionnaire was available in the electronic health record (EHR) in English for patient completion during office visits beginning in July 2020 during the baseline data collection period. The survey was completed inconsistently by providers, social work, pharmacy, and the NP during transition teaching.

Intervention

Improvement of transition readiness assessment began in January 2021. A key driver diagram (Figure 2) was created to identify interventions to improve transition readiness assessment completion. Key drivers identified included the screening method, the process of administration, provider buy-in, patient and family buy-in and participation, education and awareness of resources, and time. Two Plan Do Study Act (PDSA) cycles occurred to improve questionnaire completion which utilized technology. In the first PDSA cycle, the questionnaire was automatically deployed on electronic tablets and given to appropriate patients at visit check in. The intervention leveraged existing practices with other patient surveys being completed electronically. The process required a clinic staff member to assign the questionnaire and provide the patient with the electronic tablet. In the second PDSA cycle, the questionnaires were automatically assigned to the patients with other intake questionnaires. They could be completed via the EHR patient portal prior to the visit with other questionnaires or were automatically loaded onto the tablets which all patients receive at check-in for a visit. We were able to achieve this by working closely with our hospital's informatics team and utilizing our dual trained informatics physician as a liaison. She put in the

Transition Readiness Assessment	
Survey Questions	**Answer choices**
General Questions	
Who completed this form?	1 - Patient 2 - Parent 3- Other
How important is it to you to prepare for/change to an adult doctor before age 22?	0-10
How confident do you feel about your ability to prepare for/change to an adult doctor?	0-10
First Steps Stage: Learning about my healthcare	
I can explain my medical needs to others	Yes No, but I am ready to work on this No and I am not ready to work on this N/A
I know my allergies to medicines	
I know my own medicines, when, and how to use them	
Understanding Stage: Leraning how to manage my health care	
I know when I turn 18, I have full privacy in my health care	Yes No, but I am ready to work on this No and I am not ready to work on this N/A
I know how to refill my medications when I need to	
I know how to use MyChart to find my important medical information	
Beginning Independence Stage: Managing my health care needs on my own	
I know how to make and cancel my own appointments	Yes No, but I am ready to work on this No and I am not ready to work on this N/A
I know what health insurance I have and carry it with me every day along with other important health information (eg insurance card, emergency conbtact, etc.)	
Adult Care Ready Stage: Almost ready for transfer	
I know when and how to get medical care when my provider's office is closed	Yes No, but I am ready to work on this No and I am not ready to work on this N/A
I know what health problems my family members have	

FIGURE 1
The questionnaire is 13 questions. The first column shows the questions, and the second column shows the potential answer choices. Please note that for questions 4-13, each question's answer choice is the same and includes the following options: yes; no, but I am ready to work on this; no, and I am not ready to work on this; N/A.

EHR build request via online form for the pre-existing survey to be loaded automatically onto the tablets. The survey had already been built in the EHR during the hospital wide initiative the year prior.

Study of interventions

The percentage of patients with transition readiness assessments completed was measured each month in a statistical process control chart. Partial responses were counted. The transition QI team met monthly to review data and plan future interventions. A QI specialist assured the quality of the data and validated reports received from the hospital informatics team.

Measures

Our main outcome measure as stated above was the percentage of patients with transition readiness assessments completed each month. Specifically, our numerator was the number of patients completing the transition readiness assessment each month and our denominator was the number of patients aged 14 and up who had been seen at least 3 times in rheumatology clinic and had a visit during that month. Additional process measures included percentage of questionnaires completed by patients (vs. parents/caregivers), and beginning in March 2023, percentage of questionnaires with all questions answered (i.e., tracking partial vs. complete survey completion). A balancing measure included the percentage of patients > age 21 seen in the past year to monitor if questionnaire completion and focus on transition readiness caused patients to transition later. While reviewing our data later in the stages of the project, it was noted that an incorrect denominator was being used that did not consider specific visit types including some of our specialty clinics, multidisciplinary clinics, and video visits. Therefore, the data was retroactively corrected beginning in April 2022 to add these visit types and more accurately represent our transitioning population. We also realized that the previous metric analyzing the percentage of

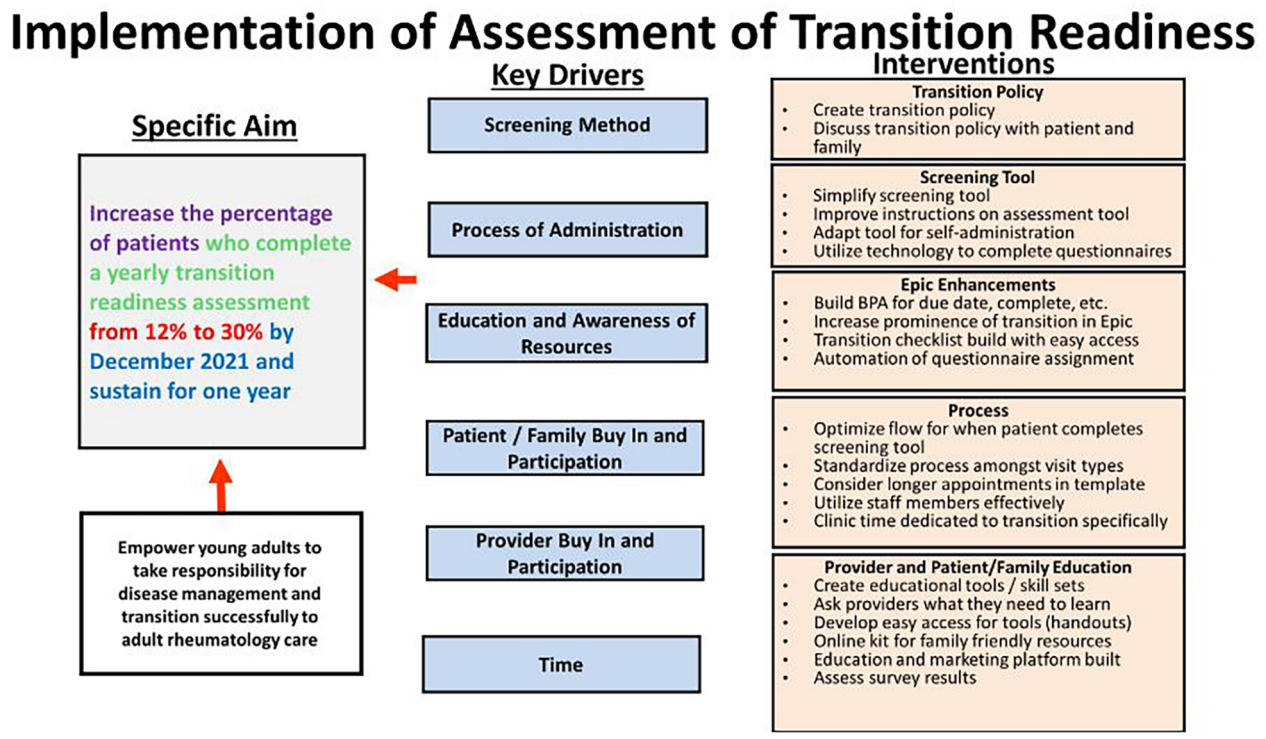

FIGURE 2
Key driver diagram.

patients who completed the assessment of all patients in the target population was flawed as this did not factor in the EHR automation that only prompted patients to complete the assessment once every 12 months. Following these discoveries, we reformatted our metric and control chart with the numerator defined as the number of patients who completed the transition readiness assessment in the last 12 months and the denominator as all patients 14 and older seen in rheumatology clinic at least 3 times for specified visit types within a given month. The entire division was updated on the project's progress and familiarized with where to access survey results within the EHR 1–2 times per year.

Analysis

Questionnaire completion rates were collected from June 2019 to September 2023 with the baseline data period occurring from June 2019 to January 2021. A control chart was used to track progress of measures and evaluate the impact of interventions over time. In September 2023, we began to assess the quality of our patients' survey responses to detect self-identified transition knowledge gaps at a population level by reviewing data collected from April 2022-September 2023. Using descriptive statistics, we looked at the percentage of questionnaires completed by patients vs. caregivers as well as self-management gaps. The self-management gaps were organized into a pareto chart to identify the most common gaps.

Ethical considerations

Per institutional policy, this QI project was not considered human-subjects research. An Institutional Review Board (IRB) waiver was obtained.

Results

In total, 1,265 questionnaires were administered to 1,158 distinct patients; 74% were female and 97% spoke English as their primary language. Our baseline percentage of patients with transition readiness questionnaires completed was 12% following introduction of the questionnaire into the EHR. This event, while during the baseline data collection period, did result in special cause variation; the baseline percentage of patients with transition readiness questionnaires prior to this was 3%. Special cause variation began to occur in September 2021, prior to our first PDSA cycle, however the data continued to rapidly change so the center line did not shift until December 2021 correlating with our first PDSA cycle. The percentage of patients with transition readiness questionnaires completed rose to 95% initially with a slight decline to 92% in December 2022. As this shift occurred, our second intervention, automation of the questionnaire, was implemented and our center line remained steady (Figure 3). This completion rate was sustained for 18 months. The median age of patients completing the survey was

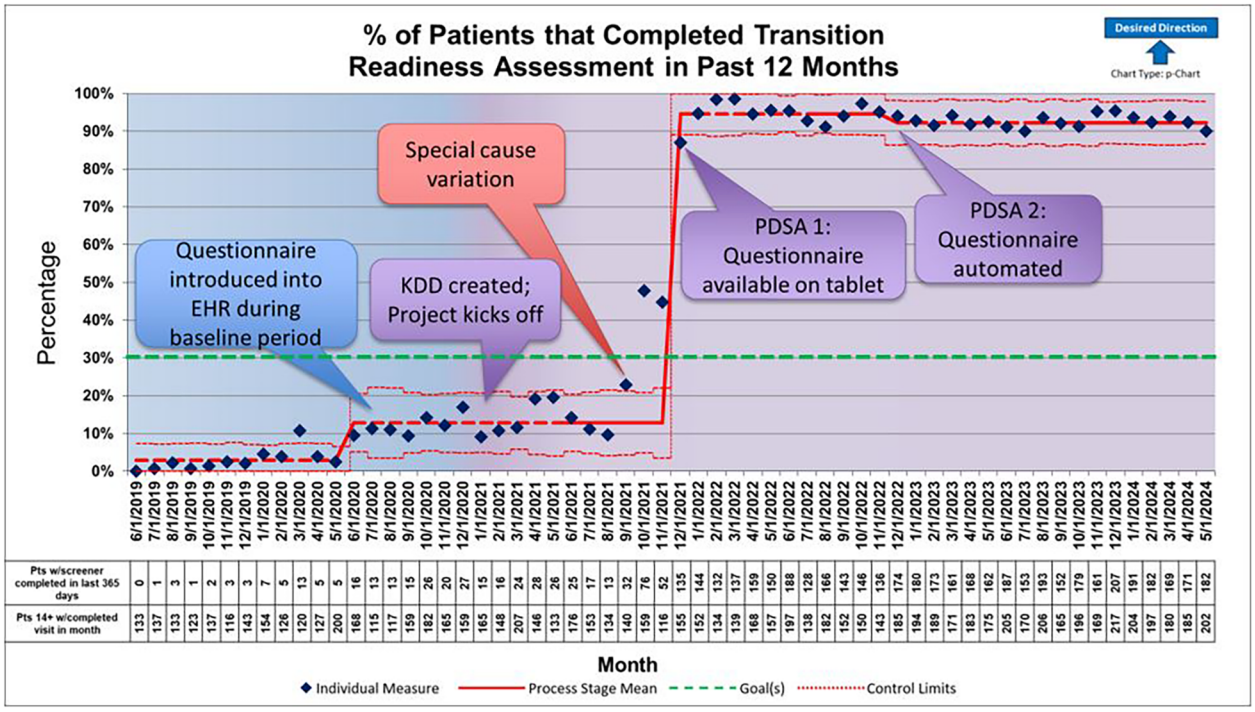

FIGURE 3
This control chart shows the percentage of patients with a completed transition readiness assessment in the past 12 months reported monthly from June 2019 until May 2024. Baseline data collection shown in blue and active quality improvement initiative shown in purple beginning in January 2021. The first Plan Do Act Study (PDSA) cycle occured in December 2021 and the second PDSA cycle occured in December 2022. Special cause variation was seen in September 2021 with a centerline shift in December 2021 after data points stabilized to a mean of 95%. It decreased slightly to 92% at the time of our second PDSA cycle in December 2022 and was sustained. Special cause variation was also seen in June 2020 during baseline data collection period.

17 years old. Fewer than half of 14-year-old patients completed the questionnaire themselves; the patient completion rate increased to age 20–21, then decreased (Supplementary Figure S1). The most frequently identified educational needs were related to health insurance, scheduling appointments, obtaining care outside of rheumatology clinic business hours, EHR messaging, and refilling medications. These five factors accounted for more than 80% of the identified needs (Figure 4). Our balancing measure of percentage of patients greater than age 21 seen during each month decreased slightly from 3.5% to 3% during the project.

Discussion

Using QI methodology and clinical informatics principles, we were able to successfully create a sustainable model for tracking transition readiness through automation of survey dissemination to our AYA patients yearly. We have achieved greater than 90% annual completion of transition readiness assessment. Our first PDSA cycle, electronic survey implementation, seemed to contribute most to the rise in completion rates though the special cause variation preceded the PDSA cycle by 3 months. This likely occurred due to increased focus on the questionnaire during this time. However, removing human factors such as the need for survey assignment improved sustained effect. The automation of the process is a strength of the project as it facilitates lasting sustainability (15).

Our findings are in alignment with other QI work within pediatric rheumatology which identify that automation within the EHR is an important facilitator of successful implementation of healthcare transition processes (16). This approach has allowed our efforts to sustain and collect data from most of our transitioning AYA. We will continue to use this approach as our efforts shift into the project's next phase. Several studies have looked at evaluating transition readiness with questionnaires, but few have taken a QI lens to improve implementation and only one has looked at questionnaire scores longitudinally (10). Interestingly, this study, which followed patient scores over time in a specialty clinic and did not have a formalized transition process, found that baseline scores did not predict transition or time to transition (10). It remains to be seen if addressing targetable skills will improve transition outcomes; however, self-efficacy, resilience, and patient activation have been shown to predict transition readiness scores suggesting that focusing on self-management skills and patient empowerment can improve scores and hopefully promote successful transition (17–20). While this is only one small step in a successful transition program, we believe that creating reliable assessment of transition readiness will allow future interventions to be more impactful.

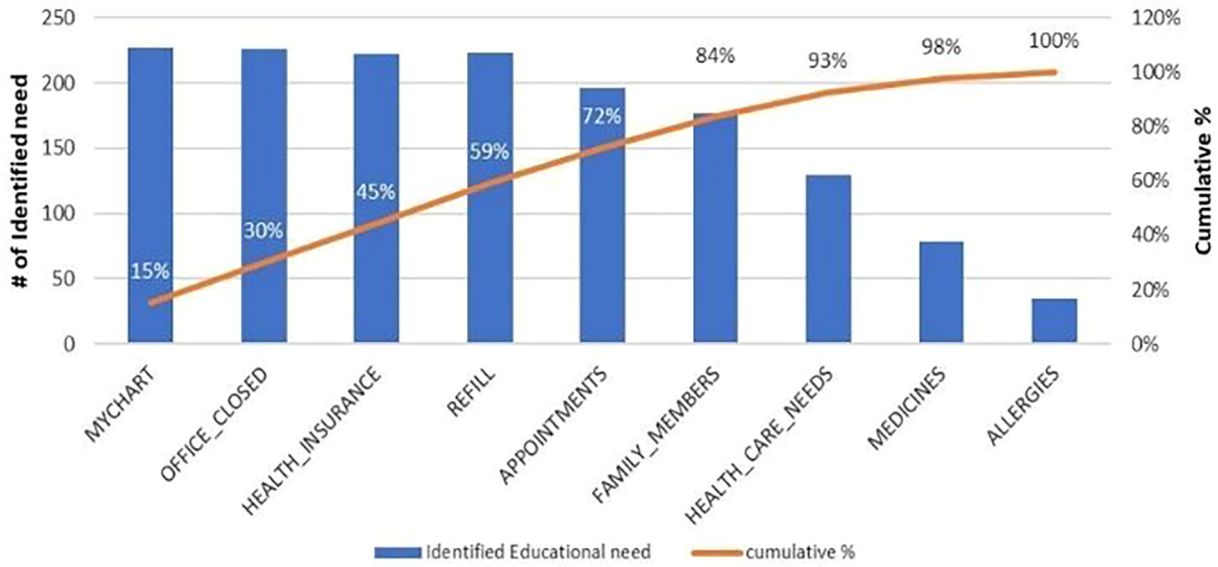

FIGURE 4
This pareto chart represents the most identified edicational needs based on data from the transition readiness assessment responses. Knowledge of patient portal use, what to do when the office is closed, knowledge of personal health insurance, understanding how to refill medication and kowning how to schedule and cancel appointments accounted for more than 80% of identified needs.

As previously noted, our outcome measure was being inappropriately captured due to a false denominator. Fortunately, we have since adjusted this metric and our corrected completion rates are consistently greater than 90%. Additionally, we observed that a proportion of surveys were completed by parents, so results may inaccurately reflect the patients' educational needs and transition readiness and instead reflect the parents' perceptions. Our current process also does not distinguish between partial and total completion of questionnaires. We also do not have surveys for non-English speaking families which is a critical population that may be more vulnerable to unsuccessful transition.

We found it interesting that survey completion by patients increased only to age 21 and then decreased. However, when reflecting on the population of patients we serve greater than age 21, we note that many of them have intellectual disability. Many of them are unable to complete any questionnaire independently and thus parents are likely completing the questionnaires more in this age group.

While these issues are all current limitations of this project, we plan to address them in future iterations. Future directions will focus on improving the quality of the data and beginning to target educational interventions. To improve data quality, we plan to improve the percentage of questionnaires completed by patients, redefine survey completion to include all questions, measure incomplete survey completion as a process measure, and provide the survey to the most common non-English primary languages seen in our clinic. These changes should improve the utility of survey responses.

Survey responses are all viewable in our EHR in a specialty specific tab. Currently, there is no specific process in place to review these questionnaires with patients. It is up to the provider to review and discuss these responses and provide targeted education focused on improving patient knowledge and skill acquisition as needed. Our QI team is currently focusing on interventions to standardize this process. As a first step, we have started monitoring percentages of "yes" responses with a breakdown by age group for targeted and prioritized educational interventions. We will start by targeting the most desired educational needs including health insurance, scheduling, and medication refills identified in our pareto chart. The QI team just completed a brainstorming exercise and then created an effort/impact matrix to identify educational interventions and determine our next PDSA cycles.

Given the complexities of transition, QI efforts lend themselves to improving this challenging process. Even with a robust team and informatics support, this initiative took over 1 year to successfully implement and is only a small step in the overall goal of a comprehensive transition of care program. We hope this approach will encourage other centers to try to progress in their transition efforts. Our institution is fortunate to have many resources which help move this project along. However, even with more limited resources, utilizing a QI framework can help center transition efforts, and there are ongoing efforts to integrate more transition content into EHRs to facilitate this process. Based off our experience, we recommend the following:

- Start by formulating a QI transition team that meets at a regular interval. Measuring transition readiness can start out simply with paper survey administration.

- It is useful to leverage resources that institutions may have.
  - If an institution has any QI team, utilize them as much as able, particularly for data collection.
  - Find champions within the division, and utilize support staff such as nurses, social workers, and allied health professionals.
  - Leverage existing practices in the division. If surveys are already being administered for other things, add a transition readiness assessment to that process.
  - If automating or integrating into the EHR, utilize an informatics team.
- Meet regularly and expect delays. This process is not linear, and setbacks are normal and to be expected.

In conclusion, this initiative allowed us to identify modifiable knowledge gaps deemed necessary for successful transition on both a population and individual patient level. We will target these areas for knowledge acquisition in the next phase of our larger transition of care QI initiative.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The requirement of ethical approval was waived by Nationwide Children's Hospital Institutional Review Board for the studies involving humans because this is a quality improvement project. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin because this is a quality improvement project and surveys are part of clinical care.

Author contributions

MA: Investigation, Methodology, Project administration, Writing – original draft, Writing – review & editing. EM: Data curation, Formal Analysis, Writing – review & editing. AT: Conceptualization, Data curation, Project administration, Resources, Software, Writing – review & editing. KW: Project administration, Writing – review & editing. PJ: Conceptualization, Writing – review & editing. AG-L: Project administration, Writing – review & editing. BT: Project administration, Writing – review & editing. AS: Project administration, Writing – review & editing. JG: Data curation, Formal Analysis, Methodology, Project administration, Writing – review & editing. AL: Data curation, Formal Analysis, Investigation, Methodology, Project administration, Validation, Writing – original draft, Writing – review & editing. SA: Conceptualization, Investigation, Project administration, Supervision, Writing – review & editing. VS: Methodology, Project administration, Supervision, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2024.1457651/full#supplementary-material

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Key data elements for a successful pediatric rheumatology virtual visit: a survey within the PR-COIN network

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Introduction: Juvenile idiopathic arthritis (JIA) is the most common childhood rheumatic disease which is commonly monitored by a combination of history, physical examination, bloodwork, and imaging. The COVID-19 pandemic prompted a rapid shift to telemedicine to ensure that patients continued to receive healthcare. The shift to telemedicine changed the methodology and ability of healthcare providers to monitor their patients' progress, as they were unable to perform direct hands-on assessments. The following survey sought to understand the impact of switching pediatric rheumatology healthcare delivery from in-person to telemedicine modality. Specifically, it sought to examine the rate of collection of critical data elements (CDE) for monitoring JIA disease activity and outcomes, barriers and facilitators to its collection, opinions on difficulty and importance of collecting CDE over telemedicine, tools and electronic medical record modifications that facilitated CDE collection, and other data elements that were important to collect during telemedicine visits.

Methods: A cross-sectional survey was sent to healthcare providers at all PR-COIN centers who saw patients using telemedicine. Qualitative data was analyzed using descriptive statistics and qualitative data was analyzed using an inductive approach.

Results: Survey respondents reported that they documented the CDE at least 75% of the time. Barriers to assessing and documenting critical data elements included (1) the inability to palpate or visualize all joints over telemedicine, (2) connectivity issues, and (3) forgetfulness with collecting all CDE. Respondents suggested using reminders within the electronic medical record to prompt documentation completeness and improve reliability. They also suggested

including medication adherence, quality of life, and patient/caregiver satisfaction with their telemedicine experience as part of their documentation. A few centers reported that they had established processes to assist with data collection in advance of the telemedicine visit; however, the variation in responses reflects the need to standardize the process of providing care over telemedicine.

Discussion: Multiple barriers and facilitators to collecting CDE during telemedicine visits exist. Given that a proportion of the population will continue to be seen over telemedicine, teams need to adapt their practices to consistently provide high-quality care over virtual platforms, ensuring that patients at any institution receive a standardized level of service.

KEYWORDS

pediatric rheumatology, telemedicine, virtual, data documentation, eHealth, telehealth, telerheumatology, quality of care

Introduction

Juvenile idiopathic arthritis (JIA) is a rare, childhood chronic condition which is estimated to affect between 2 and 8 million children worldwide (1, 2). Although JIA can be effectively managed with advanced anti-rheumatic therapies, ineffective treatment can result in pain, disability, and potential vision loss from uveitis (3, 4). Healthcare providers document various indicators to monitor JIA disease activity (5–10). These may include active joint count and provider global assessment (PGA) of disease activity. Patient reported indicators are also documented including pain scores and patient global assessment (PtGA) (11). Ultimately, reliable collection of these metrics influence disease monitoring and management, thereby impacting patients' long-term outcomes.

The Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) learning network, currently comprised of 23 medical centers and parents across the United States and Canada, works collaboratively to identify and close gaps in healthcare for patients with JIA (12). They employ a "treat-to-target" strategy based on outcomes reported by both healthcare providers and patients or families is used to optimize care (13). PR-COIN previously established a set of quality measures to improve the care of children with JIA (10). Twenty measures including 10 outcome measures, 5 process measures, 4 data measures, and 1 balancing measure were included (10). Of the 20 measures identified, six were designated critical data elements (CDE): morning stiffness, joint pain, number of active joints, uveitis screening, PtGA, PGA of disease activity, which were deemed important for monitoring JIA disease activity and outcomes (10, 13). Consistent documentation and tracking of these CDE have enabled healthcare providers at PR-COIN sites to monitor their patient outcomes (10, 13). Monitoring of CDE has enabled healthcare providers to improve the outcomes of patients with JIA (14).

Access to care is essential for careful monitoring and timely management of JIA. Access to pediatric rheumatology care has long been a challenge due to the limited workforce in this field (15–17). The COVID-19 pandemic and its calls for physical distancing and quarantine further exacerbated the already limited access to healthcare providers and services (18, 19). During the pandemic, telemedicine use rose and became an alternative or complementary visit type to traditional in-person visits (20, 21). Coordinated design, evaluation, testing, adaptation, and sharing of best practices across rheumatology clinics is essential to optimize the care provided to patients with JIA in telemedicine settings (22–24).

Evidence supporting the provision of care using telemedicine in rheumatology in both the adult and pediatric populations has existed prior to the COVID-19 pandemic, but its adoption increased out of necessity for continued provision of care during the COVID crisis (25–30). An initial survey estimated that three-quarters of PR-COIN sites did not utilize telemedicine prior to the pandemic but were subsequently able to implement telemedicine by March 2020 (18). Providers felt that about half of their population could be safely and effectively seen over telemedicine (18). Although these centers were able to adapt to providing healthcare over telemedicine in the short-term, providers expressed concerns about the long-term effects of utilizing virtual care (18). This finding was not surprising given the hands-on examination is central to the examination process of pediatric rheumatology. During the pandemic, many PR-COIN site providers adopted the use of the Pediatric Gait, Arms, Legs, and Spine (PGALS) exam as an alternative to the hands-on exam (18, 31).

Recognizing the challenges of performing active joint count assessments over telemedicine, we wondered whether the shift of healthcare delivery to a virtual setting affected healthcare providers' ability to reliably collect all six CDE (10). We therefore sought to understand the healthcare providers' perspectives on the completion rates, barriers and facilitators to collecting CDE over telemedicine, which are important to successfully monitoring JIA disease activity and outcomes. The ultimate goal was to use these findings to design interventions to reduce these barriers, in turn, enabling more reliable collection of CDE via telemedicine, thereby improving the quality of healthcare provided over telemedicine to patients with JIA over telemedicine, resulting in better long-term outcomes.

Materials and methods

A cross-sectional electronic survey was created by the PR-COIN Digital-Health workgroup to characterize healthcare providers' experiences with the collection and documentation of CDE during telemedicine visits. The survey asked respondents to indicate which CDE they collected during telemedicine visits; their comfort level of collecting CDE over telemedicine; barriers to collecting CDE over telemedicine; tools that facilitated CDE collection; indicate which of the six CDE was most important to capture; which CDE was most difficult to capture, and what other data elements they thought was worth capturing during telemedicine visits. These responses were based on respondents' active recall and not an actual audit. Finally, respondents were asked to share changes which they instituted or had planned for their site's electronic medical record system as a result of delivering care over telemedicine.

A link to the voluntary, anonymous survey was sent to the lead principal investigators (PIs) of the 21 PR-COIN centers (number of existing center at the time of the study). The PIs were requested to share the survey link to their center's clinical staff who saw patients with JIA using telemedicine. The PIs were asked to confirm the number of recipients who they had sent the survey to in order to determine the denominator. This strategy was employed to avoid sending the survey to an outdated member mailing list. Participants provided implied consent to participate in the survey. The survey, which was conducted from August-September 2020.

The survey data was collected and managed using Research Electronic Data Capture (REDCap) (32, 33). REDCap is a workflow methodology and software solution designed for rapid development and deployment of electronic data capture tools to support clinical and translational research (32, 33).

Quantitative results were analyzed using descriptive statistics, and qualitative results were thematically analyzed using an inductive approach.

The PR-COIN registry and network-related collaborative quality improvement activities, including member surveys that are used as part of continuing quality improvement, were approved by Cincinnati Children's Medical Health Center's Institutional Review Board (IRB).

Results

Survey distribution and response rate

The survey was sent to the lead contact at 21 PR-COIN sites in the United States and Canada. Nineteen of 21 PR-COIN sites were represented in the survey response. Some sites were solely comprised of pediatric rheumatologists, while other sites were composed of a multidisciplinary team which included fellows and practitioners (medical professionals who are not physicians but have received additional training and are qualified to perform many similar functions as a physician, such as prescribing medications, diagnosing, treating, and managing patient care). Teams ranged from two staff pediatric rheumatologists at the smallest site to 14 staff pediatric rheumatologists, fellows, and practitioners. Fourteen (73.7%) sites reported having more than five pediatric rheumatologists at their sites.

The survey was sent to a total of 121 clinical staff who saw patients with JIA using telemedicine. A total of 119 (98.3%) completed the survey. Of the responses received, 103/119 (86.6%) surveys were fully completed, while 16 were partially completed.

Eighty-two (68.9%) respondents indicated they were staff pediatric rheumatologists, 24 (20.2%) were fellows, and the remainder (10.9%) were practitioners.

Collection and level of comfort collecting critical data elements during telemedicine visits

Respondents indicated that the six CDE data elements were collected more than half of the time (Table 1). The most documented CDE over telemedicine was morning stiffness 104/119 (87.4%), while the least commonly documented was arthritis-related pain score 77/119 (64.7%) (Table 1). Only one (0.8%) respondent indicated that they did not collect any of the 6 CDE identified by PR-COIN.

Of the 104 individuals indicating that they documented morning stiffness during telemedicine visits, 51 (49.0%) respondents indicated that they documented this parameter at every visit (Table 2). 89/104 (85.6%) respondents indicated that they were extremely comfortable documenting morning stiffness during telemedicine visits (Figure 1).

Of the 98 individuals indicating that they documented uveitis screening during telemedicine visits, 33 (33.7%) respondents indicated that they documented this parameter at every visit (Table 2). 75/98 (76.5%) respondents indicated that they were extremely comfortable documenting uveitis screening during telemedicine visits (Figure 1).

Of the 90 individuals indicating that they documented PGA during telemedicine visits, 25 (27.8%) respondents indicated that they documented this parameter at every visit (Table 2). 27/89 (30.3%) respondents indicated that they were extremely comfortable documenting PGA during telemedicine visits (Figure 1).

Of the 88 individuals indicating that they documented active joint count during telemedicine visits, 26 (29.5%) respondents

TABLE 1 Collection of critical data elements by respondents.

Critical data element	Count	(%)
Morning stiffness	104/119	(87.4%)
Completion of uveitis screening as per recommendations	98/119	(82.4%)
Provider global assessment	90/119	(75.6%)
Active joint count	88/119	(73.9%)
Patient global assessment	78/119	(65.5%)
Arthritis-related pain score	77/119	(64.7%)
Do not collect any critical data elements	1/119	(0.8%)

TABLE 2 Percentage of time critical data element collected by respondents.

	Active joint count	Provider global assessment	Patient global assessment	Morning stiffness	Uveitis screening results	Arthritis-related pain score
Never	0	0	0	0	0	0
Rarely, in less than 10% of the chances when I could have	0	2 (2.2%)	1 (1.2%)	1 (0.9%)	0	3 (3.9%)
Occasionally, in about 30% of the chances when I could have	3 (3.4%)	5 (5.6%)	5 (6.4%)	3 (2.9%)	0	1 (1.3%)
Sometimes, in about 50% of the chances when I could have	6 (6.8%)	8 (8.9%)	10 (12.8%)	2 (1.9%)	3 (3.1%)	10 (13.0%)
Frequently, in about 70% of the chances when I could have	22 (25.0%)	18 (20.0%)	18 (23.1%)	6 (5.8%)	21 (21.4%)	13 (16.9%)
Usually, in about 90% of the chances I could have	31 (35.2%)	32 (35.6%)	25 (32.1%)	41 (39.4%)	41 (41.8%)	28 (36.4%)
Every time	26 (29.5%)	25 (27.8%)	14 (17.9%)	51 (49.0%)	33 (33.7%)	22 (28.6%)

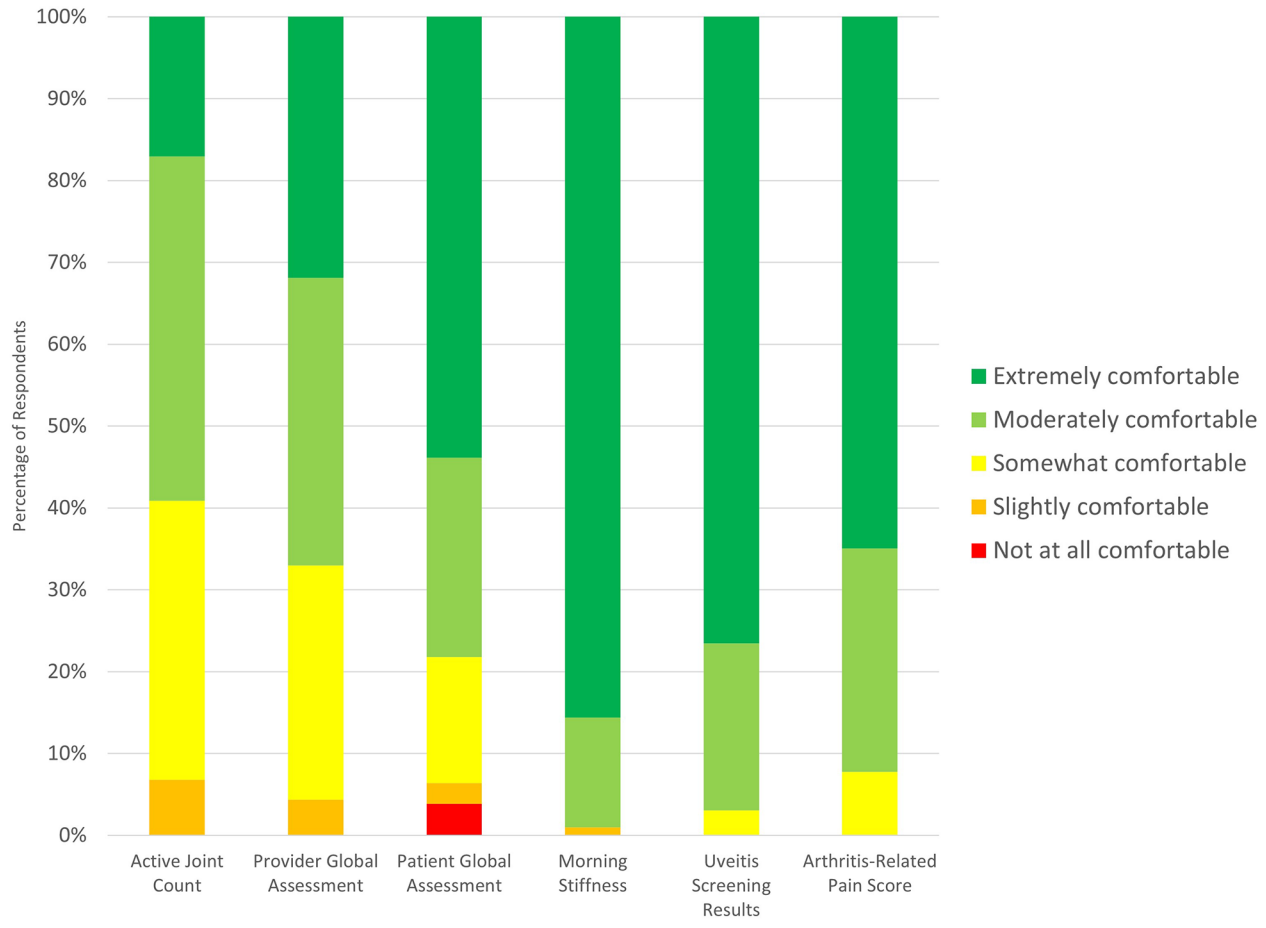

FIGURE 1
Comfort collecting critical data elements over telemedicine.

indicated that they documented this parameter at every visit (Table 2). 15/88 (17.0%) respondents indicated that they were extremely comfortable documenting active joint count during telemedicine visits (Figure 1).

Of the 78 individuals indicating that they documented PtGA during telemedicine visits, 14 (17.9%) respondents indicated that they documented this parameter at every visit (Table 2). 42/78 (53.8%) respondents indicated that they were extremely comfortable documenting PtGA during telemedicine visits (Figure 1).

Of the 77 individuals indicating that they documented arthritis-related pain scores during telemedicine visits, 22 (28.6%)

TABLE 3 Rank order of importance of critical data elements.

	Active joint count	Provider global assessment	Patient global assessment	Morning stiffness	Uveitis screening results	Arthritis-related pain
1	74 (71.8%)	16 (15.5%)	6 (5.8%)	4 (3.8%)	1 (1.0%)	2 (1.9%)
2	14 (13.6%)	31 (30.1%)	10 (9.7%)	25 (24.0%)	8 (7.7%)	16 (15.2%)
3	8 (7.8%)	21 (20.4%)	24 (23.3%)	21 (20.2%)	19 (18.3%)	10 (9.5%)
4	2 (1.9%)	14 (13.6%)	22 (21.4%)	24 (23.1%)	24 (23.1%)	18 (17.1%)
5	2 (1.9%)	15 (14.6%)	21 (20.4%)	19 (18.3%)	17 (16.3%)	31 (29.5%)
6	3 (2.9%)	6 (5.8%)	20 (19.4%)	11 (10.6%)	35 (33.7%)	28 (26.7%)

1 = Most important, 6 = Least important.

respondents indicated that they documented this parameter at every visit (Table 2). 50/77 (64.9%) respondents indicated that they were extremely comfortable documenting arthritis-related pain scores during telemedicine visits (Figure 1).

Overall, respondents appeared to be more comfortable collecting data which were reported by patients than data based on their assessment over telemedicine.

Barriers to collection of critical data elements

Forgetfulness and not knowing which tool to use to collect data were barriers for the collection of all CDE. Barriers to collecting uveitis screening results included that patients did not have their last screening date readily available during their visit nor did they have their results. Other barriers to not collecting PGA and active joint CDE included the inability to see and palpate joints and being too distracted with technical issues of using telemedicine. Additional barriers for active joints collection included difficulty assessing small joints or detecting subtle swelling, difficulty assessing young patients, and being too distracted with technical issues. PtGA collection barriers included not having the proper resources to facilitate its collection over telemedicine and an element not typically collected by a specific site. Other barriers to collecting arthritis-related pain scores included lack of proper resources to facilitate its collection over telemedicine and patients lack clarity in knowing whether their pain was related to arthritis.

Tools used to assist with collection of critical data elements

When asked what tools clinicians were using to collect CDE, the respondents from 16/19 (84.2%) centers indicated that they were using the pGALS to support the evaluation of joints. 9/19 (47.4%) centers reported that they had developed or had an existing mechanism to collect patient reported outcomes prior to the telemedicine clinic visit. 13/19 (68.4%) sites reported that they had existing reminders (e.g., forms/templates/flowsheets) or had created reminders in their electronic medical record system to remind them to collect CDE.

TABLE 4 Select the most difficult critical data element to collect over telemedicine.

Active joint count	79 (75.2%)
Patient global assessment	12 (11.4%)
Provider global assessment	5 (4.8%)
Arthritis-related pain	5 (4.8%)
Uveitis screening results	4 (3.8%)
Morning stiffness	0

Ranking of critical data elements by importance and assessment difficulty

When respondents were asked to rank which CDE they thought was most important of the six, the majority indicated that was the active joint count 74/103 (71.8%) (Table 3).

When respondents were asked to select which was the most difficult of CDE to collect during the telemedicine visit, the majority 79/109 (72.5%) indicated that it was the active joint count (Table 4).

Other elements to collect during telemedicine visits

When survey respondents were invited to suggest additional elements worth collecting during telemedicine visits, the majority suggested collecting a satisfaction survey regarding patient's telemedicine experience. Other suggestions included medication adherence, mood assessment, limitations in activities of daily living, quality of life, and the number of non-billable encounters that occurred over telemedicine.

Modifications to electronic medical record system to delineate telemedicine visits

The majority [60/109 (55.0%)] of respondents indicated that their site had made changes to their electronic medical record system to indicate that visits were conducted over telemedicine. 22/109 (20.2%) respondents indicated that their electronic medical record system already had the capability of distinguishing which visits were conducted in-person and which visits were conducted over telemedicine. Eight (7.3%) respondents indicated that their site intended to make changes to their electronic medical record

system in the future to enable them to distinguish which visits occurred in-person vs. over telemedicine.

Discussion

The COVID-19 pandemic has resulted in significant changes in healthcare delivery in both the inpatient and ambulatory settings (34–36). For pediatric rheumatologists, this change has been most apparent in the outpatient setting given that many patients with chronic disease, including JIA, require frequent outpatient follow-up visits. Although the availability of telemedicine increases access for our patients (26, 37), we must consider not only access and acceptability, but also the quality of healthcare delivered over this medium, which may ultimately affect safety and patient outcomes (38). Our initial work (18) indicated that there was a significant variability in the reliable collection of many data elements needed for clinical care at a PR-COIN site level. This study focused on individual provider practices. We observed that the majority of providers were collecting CDE at least 60% of the time when seeing patients over telemedicine. Certain CDE were collected more reliably than others. This may have been related to similarities in how the CDE is administered during in person visits. For example, morning stiffness is often verbally asked of the patient or proxy during their in-person clinic visits.

The inability to perform hands-on physical examinations mostly affected provider's ability to determine active joint count and, in turn, the PGA. This uncertainty, in turn, made them less comfortable in documenting their findings into the patient's electronic medical record.

Morning stiffness and uveitis screening were the most commonly collected CDE. However, when providers were asked to rank the importance of these elements, they considered these elements less important compared to arthritis-related pain score and PtGA. This indicates that although providers were collecting some data elements, not all elements were reliably collected.

Positive experiences and acceptability have been reported by the majority of patients/caregivers, especially when considering factors like the distance of patients' residence from the healthcare provider, patients' educational level and the perceived benefits for social distancing (39, 40). In addition to reduced travel time, decreased missed time from work/school and financial savings associated with in-person visits, patients reported ease of use, shorter waiting periods and possible continued use in the post-pandemic period (26, 27, 41–43). Healthcare providers also reported high satisfaction, especially when patients had reliable internet (44). Common barriers identified with practicing telemedicine include lack of physical examination, reduced diagnostic accuracy due to incomplete clinical information, difficulty reaching patients, missing nonverbal communication, and lack of or challenges using technology required for telemedicine visits (45, 46). Barriers unique to the pediatric rheumatology population include trying to keep very young patients focused during virtual physical exam, and difficulty assessing psychosocial factors in adolescents when caregivers are present (46–48). Unfortunately, lower socioeconomic status and lower educational background may affect access to and quality of telemedicine visits e.g., poor bandwidth, which has implications in continuity of care, medication adherence and disease control (49–51). The quality of virtual care may also depend on the specific disease and its activity level. A randomized controlled trial demonstrated that telemedicine visits were not inferior to in-person visits for adult patients with rheumatoid arthritis whose disease was in remission or had low disease activity (52).

Although barriers to data collection were in part due to the nature of telemedicine and limitations in exam, a large contributor was simply due to provider workflow issues. The inexperience and lack of training in using telemedicine platforms, completing virtual patient check-ins, performing physical exams in a virtual setting, and the lack of support collecting patient reported outcomes, impacted their ability to collect CDE and complete their documentation.

Further, survey results indicated that for specific elements there were two main barriers: (1) the inability to conduct a reliable joint assessment that includes direct palpation of joints (especially when patients were not present at the visit), and (2) providers forgetting to collect and document the pertinent data elements. This illustrates that although telemedicine has limitations for specific aspects to the musculoskeletal exam, there are opportunities to improve workflows to collect the non-exam dependent, patient-reported data elements such as the PtGA or pain scores. As providers continue to integrate telemedicine as part of their clinical practice, we will need to consider systematic approaches to address these barriers, such as allocating job responsibilities and establishing force functions to ensure the reliable collection of CDE.

As previously indicated, fewer providers were comfortable performing physical examination to ascertain active joint count during telemedicine visit compared to acquiring other CDE due to the possibility of limited accuracy of the results. To address this concern, some providers may consider triaging patients to determine whether they should be seen virtually over telemedicine or if they should be seen in-person. To our knowledge, there is no universal established criteria on how to triage patients for telemedicine visits. One PR-COIN site utilized a pre-COVID developed triage tool that was developed prior to the pandemic which triaged based on referring symptoms to determine the urgency, time to be seen with the highest triaged levels 1 and 2 requiring in-person visit (53). Further research is also needed to identify which patients are most suitable to be seen for virtual visits and which might be better served by in-person assessment.

Alternatively, we may consider additional tools, models of care, and/or caregiver-specific education to facilitate the reliable reporting of physical examination results, including the active joint count. For example, there are already recommended modifications to the p-GALS, known as Virtual or Video-pGALS (V-pGALS), incorporating amended or additional maneuvers added to capture needed elements more accurately (47, 54). A pilot study has demonstrated the acceptability and reliability of this tool (31). Additional research needs to be performed to

further validate the ability of the V-pGALS to perform joint assessment. This could be accomplished by performing a study where patients received a joint count over telemedicine followed by an in-person assessment shortly thereafter.

There is an opportunity to improve the collection of CDE that are not dependent on the clinical exam, such as patient-reported outcomes, over telemedicine. The introduction of new clinical workflows such as the incorporation of integrated electronic health record tools (for both providers and patients navigators), provider education with time sensitive scripting and checklists, medical staff virtual rooming protocols for medical staff, and pre-visit planning, may better support reliable collection of these metrics rather than forgetting. Enabling patients and proxies to take a proactive role in their healthcare by educating them on how to support their telemedicine visit and teaching them skill may empower them whilst improving the overall outcome of the telemedicine visit.

Despite being one of the ranked one of the most difficult CDE to collect via telemedicine, respondents indicated that active joint count was the most important CDE to collect over telemedicine. Given this opinion, additional efforts should be expended to improve the ability to accurately collect this variable. Recognizing that the varying levels of knowledge and technology literacy, educational curriculums should be carefully designed to ensure that healthcare providers possess the necessary knowledge and skillset to effectively provide care over telemedicine. Furthermore, the development of additional educational electronic tools i.e., phone applications, could improve timely access to providers.

It would be worth surveying patients to understand their opinion of healthcare delivery over telemedicine and their satisfaction with the process. Some studies have indicated that although being seen over telemedicine was preferred during the pandemic, it is not preferred after the pandemic (41, 42). Additional patient reported outcome measures/surveys could be introduced through patient portal builds in the electronic medical record.

Differentiating data that is collected by telemedicine to that from in-person visits will enable the comparison of patient outcomes to determine whether the delivery of care using telemedicine results in similar patient outcomes. This information will inform whether providing care over telemedicine is comparable to that in-person care or it may identify situations where telemedicine care is a satisfactory option.

Our study is limited by the fact that it surveyed the PR-COIN learning network. PR-COIN sites have previously collected CDE during in-person visits and they have already engrained this practice into their established workflows, practices, and culture. Therefore, these findings may be biased due to the active recall design of the survey, as well as the heightened awareness and prior collection of these data elements for clinical care. As such, these findings may not be representative of the broader pediatric rheumatology community. Broader surveys and studies involving the use of these data elements, both in in-person and virtual settings, amongst pediatric rheumatologists are required.

In addition, respondents answered questions based on their own practice. We did not inquire about the composition of their practice, such as the proportion of JIA subtypes seen in their clinic or the age range of their patient population. These factors may have influenced their responses. If their practice consisted primarily of adolescents with arthritis affecting larger joints, it may be easier to perform a virtual assessments may have been easier since because they can follow instructions, and the joint swelling would be more prominent, in contrast to a toddler with arthritis affecting small joints who is unable to follow instructions.

Although the majority of video platforms used in telemedicine have matured over time, they may vary in terms of available features and ease of use. These differences can influence the technical system requirements needed to operate the software or the user's learning curve.

It is also possible that self-reported collection of data elements may not accurately reflect actual practices, potentially over or underestimating actual practices. Collecting objective data on the frequency that these metrics are captured during visits would more definitively identify gaps. Additionally, while this survey primarily captures largely the provider experience with collecting data elements via telemedicine, future next steps may want to examine patient acceptability regarding the ways in which patient-reported outcomes are collected and utilized in telemedicine care. Ultimately, a deeper understanding of how collection of these data elements are collected and utilized, and how they affect patient clinical outcomes in JIA is needed and is currently being investigated currently underway.

It is important to remember that although it may be easy for some healthcare institutions to offer telemedicine to patients, health inequities still exist. These disparities can affect some individuals' ability to access care using this medium (21, 55). Additional steps must be taken to ensure equitable healthcare delivery using telemedicine in the future (25, 29, 56).

Conclusion

Multiple barriers and facilitators exist in the delivery of pediatric rheumatology care over telemedicine. Our findings suggest that telemedicine processes and practices vary both across different centers, as well as within individual centers. This highlights the need to standardize telemedicine visit procedures to ensure that CDE are reliably and consistently collected, irrespective of visit type. Given that a portion of patients with JIA will likely continue to be serviced over telemedicine post-pandemic, teams need to adapt and refine their existing clinical practices to continue providing high-quality care using this platform.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving humans were approved by Cincinnati Children's Hospital Medical Center. The studies were conducted in accordance with the local legislation and institutional requirements. The ethics committee/institutional review board waived the requirement of written informed consent for participation from the participants or the participants' legal guardians/next of kin because implied consent was used as the mechanism for consent for the healthcare providers who chose to complete the survey.

Author contributions

YG: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. MR: Conceptualization, Data curation, Formal Analysis, Methodology, Resources, Visualization, Writing – original draft, Writing – review & editing. SA: Writing – original draft, Writing – review & editing. RP: Conceptualization, Writing – original draft, Writing – review & editing. JH: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. DB: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. SV: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. TL: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. ST: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. FB-S: Conceptualization, Formal Analysis, Methodology, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that when the research was conducted, there was an absence of any commercial or financial relationships that could be construed as a potential conflict of interest. This research was conducted using data obtained through the Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN), collected by the physicians and providers participating in this multicenter Quality Improvement Collaborative. https://pr-coin.org/. Since the completion of this research, YG has received educational grant from Pfizer to improve the delivery of telemedicine visits for patients with JIA. Since the completion of the research, FB-S has departed her academic position at Nationwide Children's Hospital and secured employment at Amgen.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Digital health technology to support patient-centered shared decision making at point of care for juvenile idiopathic arthritis

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Despite availability of multiple FDA approved therapies, many children with juvenile idiopathic arthritis (JIA) suffer pain and disability due to uncontrolled disease. The term JIA includes a heterogeneous set of conditions unified by chronic inflammatory arthritis, collectively affecting 1:1,000 children. When reviewing treatment options with families the rheumatologist currently refers to the experience of the average patient in relatively small controlled clinical trials, to consensus-based treatment plans, or increasingly the choice is dictated by the formulary restrictions of insurance payers. The current paradigm for treatment selection does not incorporate real-world evidence of treatment effectiveness centered to the individual patients with whom decisions are to be made. Treatment decisions based on the evidence of the average patient are not optimized to reflect the unique clinical characteristics of an individual with JIA and their disease course, nor does it account for heterogeneous treatment effects. To guide treatment choices centered around each patient, we describe a novel concept of utilizing digital health technology to bring patient-centered information into shared decision-making discussions based on comparative effectiveness analysis of electronic health record or observational clinical registry data of patients with similar characteristics. The envisioned digital tool will organize and present data relevant to the individual patient and enable evidence-based individualized treatment decision making when used in a collaborative manner with the patient family and rheumatologist. Capabilities in digital health technology, data capturing, and analytical methodologies are ripe for this endeavor. This brings the concept of a learning health system directly to the point of care.

KEYWORDS

digital health technology, shared decision making, clinical decision support system, juvenile idiopathic arthritis, learning network model, registry analysis, personalized medicine

Introduction

Juvenile Idiopathic Arthritis (JIA) is an umbrella term for heterogeneous chronic inflammatory arthritic conditions of childhood onset that neither have a known etiology nor a cure. An estimated 300,000 children have a rheumatologic condition, and an estimated 80,000 children in the United States have some form of JIA. Despite the availability of multiple FDA approved therapies, JIA is a condition that remains uncontrolled for many children who suffer negative health outcomes, including chronic pain, growth disturbances and functional disability. Of the seven subtypes, one of the most difficult to control is polyarticular JIA (pJIA), characterized as having five or more inflamed joints, with features like rheumatoid arthritis in adults (1, 2). In a setting of multiple available treatment options, only about 40% of pJIA patients achieve a controlled disease state (3).

The lack of satisfactory disease control is likely multifactorial, but one known important factor that we seek to address using digital technology is the heterogeneity of treatment responses. Patients with JIA may respond to the same treatment differently, perhaps due to differing biology, comorbidities, or genetic factors, but heterogeneity of outcomes may also be due to the timing of treatment with respect to diagnosis, disease prognosis, use of concomitant medication, treatment duration or treatment adherence. We anticipate better health outcomes could be attained with reliable identification, selection, and prescription of the optimal treatment for a given patient chosen from currently available candidate treatments by accounting for heterogeneous factors in a comparative effectiveness analyses model.

The ability to select optimal treatments at time of diagnosis with inflammatory arthritis is vital as there seems to be a window of opportunity wherein the early achievement of clinical inactive disease within one year of diagnosis is a strong predictor of better long-term clinical and health related quality of life outcomes (4, 5). Thus it is important to understand and account for the role of heterogeneity of treatment effects in selecting initial JIA treatment.

Heterogeneity of treatment effects

Heterogeneity of treatment effect (HTE) refers to differential and non-random effects of treatments on individuals in a population compared to others, indicating that there are clinically relevant subgroups who may have different benefits (or lack thereof) compared to others (6). This is to be distinguished from the average treatment effect (ATE) estimated in studies, which would suggest that a treatment would have a similar effect across subgroups, or patients with heterogeneous characteristics (6). Historically, ATE can only be estimated from randomized controlled clinical trials (RCTs). Thanks to the theory and analytical development of statistical causal inference method, we now can utilize data observed from real clinical encounters to inform how different treatment approaches may compare. Current clinical decision-making in the main is based on ATE. There are multiple rationales for standardization of medical care and use of protocols to reduce variation in care, including reduced potential for medical error, decreased health inequities, and increased ability to perform comparative effectiveness studies in observational data. Therefore, clinicians, healthcare systems and researchers alike are motivated to pursue uniform treatment approaches across patients. However, treatment by protocols that do not consider prognostic factors and clinical presentation may not yield the best outcomes for individual patients, nor the population. Seeking consensus treatment plans that work for an "average" patient may not serve all patients due to heterogeneity of conditions and response. We believe a digital health technology (DHT) solution can be created to leverage comparative effectiveness analyses of relevant clinical patient information and present a data dashboard at point of care (POC) to inform a patient-centered and standardized care approach. We anticipate, with consideration of individual patient features such as subtype of JIA, duration of disease at diagnosis, serologic markers, sex, age, and response to prior treatments, and synthesizing the collective wisdom/experiences of care episodes, such a DHT could improve clinical outcomes. Variation in treatment across individuals informed by the DHT and based on HTE would be warranted.

Data sources to inform treatment decision-making

RCTs have been the primary data source used to establish the efficacy of medical treatments. However, RCTs are often relatively small for rare conditions such as JIA, and thus are limited in generating robust information on HTEs. Innovative trial designs such as pragmatic clinical trials, randomized withdrawal trials, and sequential multi-stage adaptive randomized trial (SMART) have been pursued recently. Yet the averaged treatment effect remains to be the primary quantity of estimation, due to methodologic challenges related to estimating patient-centered treatment effect.

Increasingly, sophisticated bioinformatics technology captures rich clinical information reflecting clinical decisions that were made at the point-of-care (POC) and the information that factored into the decision. The establishment of multi-center learning health networks that implement common data models for clinical data entry into a shared registry make it possible to combine data from multiple centers on a clinic population with data reflecting real-world treatment practices and patient outcomes. Compared to RCT data, a learning health network (LHN) registry that seeks the complete population representation for the purposes of quality improvement (QI), may offer more generalizable data and robust evidence to inform treatment effectiveness for heterogeneous and dynamic conditions such as JIA.

How to estimate patient-centered treatment effect

To estimate HTEs, the historical approach was to examine treatment by covariate interactions. However, this approach

requires testing multiple interaction terms, which raises multiplicity issues. When not addressed, this may lead to inflated type I error (7). Furthermore, such an approach imposes strong modeling assumptions, e.g., linear regression, which could seriously bias the effect estimates when a model is mis-specified. In addition, it is not always clear what covariates may modify the treatment effect, and how the covariates interact with treatment and among themselves. The challenges of HTEs are further complicated, due to treatment-by-indication bias, information biases, missing and/or censored data. Even in the RCT setting, estimation of HTEs is often complicated by intercurrent events such as early termination, loss-to-follow up, treatment switching, and/or use of rescue medications.

Statistical causal inference methods addressing HTEs largely fall within two categories – subgroup finding and conditional averaged treatment effect (CATE). Subgroup finding searches among the feature space defined by preselected patient characteristics such as age, sex, and disease subtype, identifying the subgroups (often a combinations of multiple features) that present distinct treatment effects than the averaged effect. This can be used to derive clinical decision rules based on simple and commonly available patient features and obtain estimates of subgroup averaged treatment effect (SATE). The CATE on the other hand, estimates effect of treatment conditional on the values of feature space, often leveraging on the semiparametric or nonparametric modeling algorithms such as random forest. Bayesian adaptive regression tree (BART) modeling is widely recognized to provide well performed ATE and has been suggested to model CATE (8). A concern with a highly flexible modeling approach is overfitting, which could lead to overly confident estimates that are not reproducible in another study sample. Setting aside a subsample of data may help achieving better "honest" inferences to the estimated treatment effect using an adapted random forest approach, where the node splitting criteria is designed to optimally create multiple subgroups of HTEs (9). When treatment-by-indication confounding bias is of concern, doubly robust causal inference methods are used to introduce additional safeguards against potential model misspecification (10–13). Bayesian Gaussian Process (GP) utilizing GP covariance function as a matching tool, can provide a Bayesian's doubly robust approach (14, 15). The Bayesian approach is well-suited for synthesizing and updating knowledge for informing evidence-based decision making. The Bayesian framework, where the prior represents the existing knowledge, uses new data to update the prior and produce the posterior that represents the updated knowledge synthesizing both past and new learning. These Bayesian approaches can explicitly consider the multiplicity issue (16), search subgroups with distinct treatment effects (17), and be coupled with nonparametric models to mitigate the model misspecification issue in HTEs (18). The decision-based Bayesian causal inference method can be used to identify patients who may experience clinical meaningful improvements from a given treatment (17).

The causal inference methods HTEs brought us much closer to better understanding patient centered treatment effect. However, much work remains to rigorously validate the HTEs provision of causal inference at the individual level to inform individual treatment effect (ITE). For example, CATE and SATE neglect the inherent variability in response measurements or due to finite samples, yet consideration of these variabilities is critical to inform decision-making. Building on the existing HTE methods, we seek to identify a better performing approach with the goal of delivering relevant and valid comparative effectiveness treatment evidence for each individual patient. Towards this goal, the chosen method to inform treatment decisions at point-of-care should meet the following criteria: (a) it provides an accurate estimate of ITE; (b) it provides nominal level of confidence in a treatment choice; and (c) it is computationally efficient and feasible at point-of-care.

To validate methods for informing ITE, we need to assess the performance of the method with an independent sample of the "target" patient with whom the decisions are to be made. We may do so by taking a leave-one-out (LOO) approach with the existing data source. However, due to the inherent variabilities in responses and sample heterogeneity, the observed outcome for the out-of-sample "target" patient is only a random realization of many versions of possible outcomes. This imposes the seemly infeasible task of performing validation for ITE, unless we have access to the expert clinicians and consensus agreement. By having access to the data recorded from the real clinical encounters of thousands of patients cared for by hundreds of physicians, we in fact do have access to the requisite expert opinions. For each individual patient sitting in the doctor's office, we could identify the subsample of patients in the database that resemble or are "alike patients". This means, the treatment decisions made and the outcomes following the corresponding decisions can be extracted. The summary statistic pooling data from all physicians could serve as an anchor point, allowing us to validate and compare the performance of different causal inference methods for informing patient centered decisions.

The top performing models can be implemented in a DHT, which will take the input of patient data in the EMR, clinical registries, and clinical trial data inputs. The DHT may also be updated with additional data information accumulated as more data is made available. The DHT should generate output that provides patient-centered estimate of treatment effect based on patient characteristics in a format designed in a manner to review and discuss with the patient.

From digital health technology to shared decision making

Clinical decision support (CDS) is a model element to improving chronic illness care (19). In rheumatology, active monitoring of disease status and medication adjustment if treatment targets are not achieved—a strategy called "treat to target" (T2T)—results in tighter disease control (20), and better long-term outcomes. Adding a CDS tool increases the impact of T2T (21), and a study of T2T with CDS in JIA suggested that use of CDS over time could potentially ameliorate racial disparities in disease activity that had been identified at diagnosis (baseline) (22). However, in clinical practice, a T2T approach in JIA is limited by the lack of evidence-based, accessible at POC CDS on

next best treatment decisions that would be expected to result in better disease control considering the patient characteristics.

Recent consensus recommendations from pediatric bioethicists stated, "to respect children and promote their wellbeing, clinicians and parents should inform pediatric patients of salient information and invite their perspective to the degree that doing so is developmentally appropriate." (23) To this end, the digital health tool that will be presented as a CDS to inform treatment decisions at POC should be designed with the intent to be both provider and patient facing to support collaborative shared decision making with patients and their care takers. The concept is illustrated in the Figure 1. The algorithm informing the digital health tool includes an age span of 1–18 years. Initial development and analysis of the tool centers on decision making with parents, but with a clinical goal of including patients in the process. Future research will study the dynamics of parent-child dyads in the decision process.

Why a patient-centered shared-decision making tool can make a difference

Human intelligence learns from what we observe and applies learnings to future decisions. Prior to the big data age, clinicians learned from published textbooks or the medical literature, their past experiences, and from communications with their peers. For each future patient, the more we accumulate past knowledge relevant to the patient, the better we are at making treatment decisions. As a result of advances in immunology, together with biotechnology innovations and modern pharmaceutical product development, the medical field has made great advancement in treating JIA disease conditions with improved health outcomes. However, treatment of JIA, a heterogeneous group of conditions, is complex, and the disease course unpredictable. Even in this increasingly rich information environment JIA treatment continues to involve guesswork and anecdote, subject to human error, bias, and unwarranted variation in care.

What if we could add a patient-centered learning algorithm into decision making at POC? Powered by such an algorithm validated in a research setting (14), we aim to build an interactive, user-friendly CDS to support shared decision-making at POC. The use of such a tool can address health equity concerns by standardizing the evidence-based approach taken with each patient. Resulting treatment variation between patients would be warranted based on computational predictive analytics of most effective treatment resulting in individualized care. Rather than "one-size-fits-all", the individualized treatment approach will consider, through algorithm learning, what has worked well in similar patients.

Application to a learning health system: development and testing

Healthcare equity is a central consideration in the design and implementation of any new DHT. The Agency for Healthcare Research and Quality has developed a Digital Healthcare Equity Framework guided by principles that the new technology should reduce inequities, be person-centered, be inclusive in development, be able to be implemented in diverse settings, be cognizant of policy, and be focused on outcomes (24). The framework details aspects of development to consider developing a DHT that promotes health equity. These recommended developmental approaches include engagement of diverse potential end-users, identification of potential cultural barriers to use to design around, when developing workflows paying attention to access to information technology, obtaining iterative feedback on whether technology is serving needs of the end-users, and inclusion of representative data in development (24). These guiding principles and domains are important to bear in mind with any healthcare delivery improvement. Usability surveys and quality measures (process, outcomes, healthcare experience) will be stratified by demographic features or social determinants of health throughout pilot testing of a new DHT and after implementation to monitor for equitable application.

A LHN is an optimal context to develop, test and deploy such a DHT. The Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) is such a LHN on a shared clinical registry populated by electronic data transfer from the EMR, local databases, or manual data entry extracted from the EMR (25). Participating sites are unified in a shared and relentless focus on improving outcomes of all JIA patients using QI methods, with attention to standardized care, avoidance and mitigation of quality-of-care gaps. Therefore, registry data are more heterogeneous than in clinical trial databases or research registries that select for specific JIA categories. Populated by pediatric rheumatology centers characterized as innovators and early adopters, with a platform to track quality measures, the network is an ideal setting to test the health equity principles outlined by AHRQ as a research prototype DHT is translated into a viable clinical tool. Qualitative research with anticipated end-users (clinicians and patients) from diverse clinic settings and backgrounds, will increase adaptability. User-centered design expertise in the iterative design and development of the tool, use of QI approaches to pilot the integration of the tool into the clinical workflow are factors that are increasing likelihood of successful future adoption. Barriers to use are anticipated with respect to integration of the DHT into the local EMR interface requiring local leadership buy-in and resources, increasing complexity of real world data to be integrated into updating treatment algorithms, time constraints of introducing new technology and presenting data to patients for shared decision making, required training of clinicians and staff, legal and regulatory requirements related to data flows, potential dependency on technology access and concern for introducing health inequities, need for cross-cultural and language translation in using the tool with languages other than English.

Discussion

Advances in use of digital technologies, including health information technologies and real-world data analytical

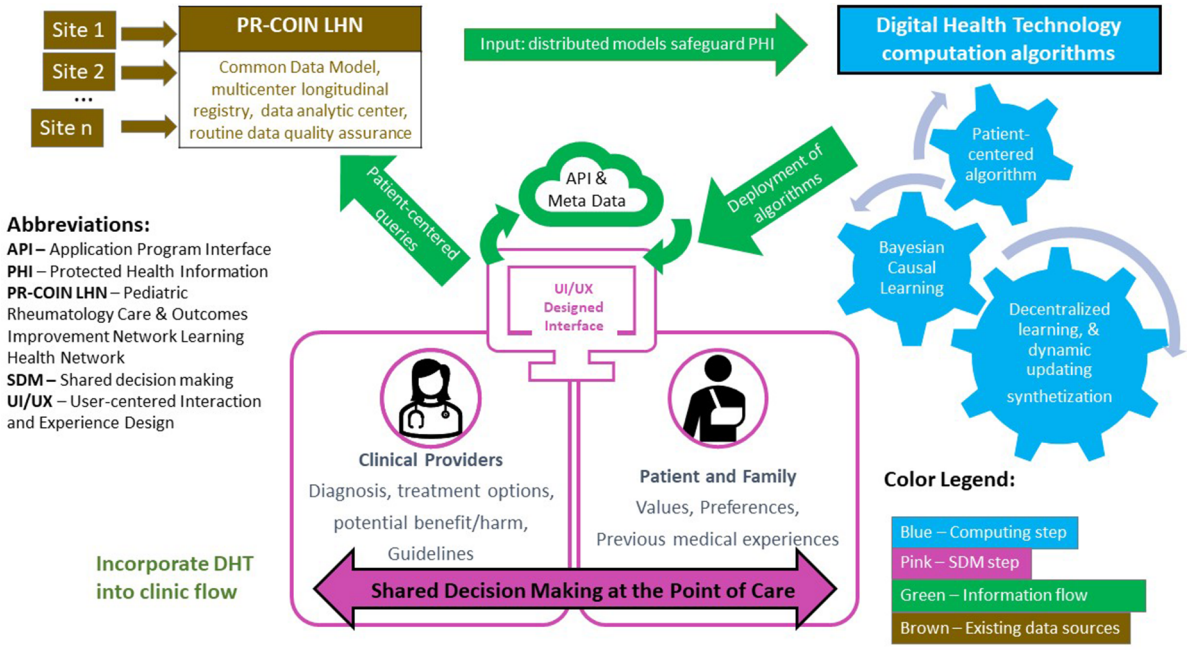

FIGURE 1
Conceptual framework: digital health technology enabled patient-centered shared decision making at point of care.

technologies, and the increased incorporation of digital devices in our daily lives, create the context and environment for digital CDS tools to be offered at POC with promise to deliver more efficient, effective patient-centered care. The increasing sophistication of EMR and real-world data captured by modern technology into registries creates the opportunity for achieving evidence-based personalized medicine. These data sources together with the appropriate methods, and emerging infrastructures hold much promise to enable patients and physicians to make shared, informed decisions tailored to an individual patient by learning from the experiences of "alike" patients.

However, real-world data can be misleading. Unlike clinical trials, patients are prescribed treatment based on their disease indication (treatment-by-indication), and patients who fail to respond may then be put on an alternative or additional treatment (post-treatment selection bias). Without carefully managing such treatment-by-indication and time-varying post-treatment selection biases using causal inference methods, we cannot obtain unbiased real-world evidence. RCTs are useful for informing population-averaged treatment but are rarely sufficient to inform patient-centered adaptive treatment effect. The causal inference methodologies addressing HTEs and the time-varying adaptive treatment strategy, are increasingly sophisticated and able to handle real world complexity, application of the method requires advanced knowledge and computation programming skill, as well as the ability to harmonize and access multiple data sources.

We envision capability for patient-centered causal learning as the engine of a new kind of smart CDS in form of a DHT. It can utilize large heterogeneous data sources and address multiple data challenges inherent in use of EHR data. The DHT will then bring patient-focused evidence on the comparative effectiveness of treatments in patients like them to the POC and thus improve patient-centered treatment choices. A DHT works well within the scope of a learning health system, which can feed data from clinical care to support shared learning and inform treatment algorithms, leverage QI approaches and a drive towards health equity in development, to test and implement the system equitably in clinical care.

Data availability statement

The data analyzed in this study is subject to the following licenses/restrictions: The datasets analyzed for this study are housed at Cincinnati Children's Hospital Medical Center on a secure server. De-identified data are accessible on reasonable request. Requests to access these datasets should be directed to bin.huang@cchmc.org.

Ethics statement

Regulatory approval for the "Bayesian Causal Inference Methods Development fpr Comparative Effectiveness Research in Patients with Chronic Conditions" study (IRB ID 2021-0242, PI Huang) is obtained from Cincinnati Children's Hospital Institution Review Board. Regulatory approval for the study "Inform Shared Decision-Making with Advanced Causal Inference to Improve Quality of Pediatric Rheumatology Care" (IRB ID STUDY00004570, PI Morgan) is obtained from Seattle Children's Research Institute.

Author contributions

BH: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. MK: Conceptualization, Formal Analysis, Investigation, Methodology, Supervision, Writing – review & editing. CC: Data curation, Formal Analysis, Writing – review & editing. ND: Methodology, Writing – review & editing. KF: Methodology, Writing – review & editing. MM: Methodology, Writing – review & editing. HB: Data curation, Writing – review & editing. DL: Conceptualization, Data curation, Methodology, Writing – review & editing. EM: Conceptualization, Funding acquisition, Investigation, Methodology, Project administration, Resources, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Achieving reliable patient reported outcomes collection to measure health care improvement in a learning health network: lessons from pediatric rheumatology care and outcomes improvement network

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Introduction: Data from the Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) registry suggests that reliable collection of patient-reported outcomes (PROs) varies across sites. The objective of this study was to better understand the practices of collecting PROs at PR-COIN sites.

Methods: A REDCap survey was sent to the lead representative for each PR-COIN site. Registry data were analyzed to better understand the completion rates of PROs. Interviews of physician leaders of high performing sites were conducted by videoconference, audiotranscribed and themes were summarized. Quantitative data were analyzed using descriptive statistics and qualitative data were thematically analyzed.

Results: All 23 PR-COIN sites responded to the survey. PROs were collected by 21/23 (91%) sites. Arthritis-related pain intensity, morning stiffness, and physical function were the top three collected PROs (Supplementary 3 and 4). PROs were collected using paper, electronically or in combination, with most sites collecting PROs only on paper. PROs were manually scored at most sites. Among sites with electronic PRO collection, 42% did not have automatic transfer of scores into the electronic medical record. Facilitators to successful collection of PROs included availability of staff, training, and culture. Barriers to PRO collection cited were limited time, lack of infrastructure, and lack of staff. Completion rates of PROs in the registry in top 4 performing centers for morning stiffness was 100%, overall well-being and pain intensity scores ranged from 93%–98%, and for physical function 69%–94%. Interviews with physician leaders indicated that their site overcame barriers through: integration of PRO collection into workflow, gaining buy-in of stakeholders (clinicians and patients), and automating PRO collection. Interviewees endorsed automation of data collection (e.g., self-completion on tablets) and automated transfer to electronic medical record (EMR) as key components enabling reliable PRO collection.
Conclusions: Through understanding our current ability to systematically collect PROs across all sites in PR-COIN and exploring successful implementation of PRO collection both within and outside our learning health network, we share lessons learned and identify the most influential factors for successful PRO collection in pediatric rheumatology.

KEYWORDS

patient reported outcomes, pediatric rheumatology, quality of life, outcome measures, juvenile idiopathic arthritis

Introduction

As healthcare moves towards more patient-centered care, it is imperative to examine methods of integrating patients' opinions into clinical assessments and decision-making. In pediatrics this is achieved by obtaining input from either the patient themselves or, in cases where the patient does not have the developmental capacity, their proxy. Patient-reported outcomes (PROs), defined as "any report of the status of a patient's health condition that comes directly from the patient without interpretation of the patient's response by a clinician or anyone else", are an important tool for measuring patient outcomes (1). Patient-reported outcome measures (PROMs), validated questionnaires which are used to measure PROs, are completed by patients and their proxies to inform their healthcare teams about their perception of their health status and quality of life.

Pediatric Rheumatology Care and Outcomes Improvement Network (PR-COIN) (2) is a learning health network (LHN) dedicated to improving healthcare delivery and patient outcomes through quality improvement methodology (3). Patient data across the network are collected in a central registry (4). Patient engagement is a central component of a LHN, and the patient voice is integral to care through shared decision-making. The PR-COIN LHN's focuses on outcomes improvement prioritizes disease control, relief of pain, and optimization of physical function through a "treat to target" strategy (5). Striving for complete data collection is a critical first step toward understanding disease activity status, gaps in care, and ultimately, planning impactful interventions to improve health outcomes. As this LHN is a collaborative between patients and families, the collection is PROs is important for patient-reported outcome data in the case of a LHN that is co-produced with patients and families and prioritizes outcomes that are important to patients. Qualitative research with patients and families indicate that pain, physical function and patient perception of overall well-being are outcomes they prioritize to be measured in longitudinal observational studies and clinical trials (6) and are therefore collected as quality measures in PR-COIN (3).

Despite the recognition of the importance of these health domains to patients and intent for reliable collection of these measures, collection of PROs within PR-COIN varies across sites. To better comprehend the various practices for collecting PROs within PR-COIN, the PRO Standardization Workgroup conducted a survey of sites to determine which PROs were being collected, to understand operational processes to PRO completion, and to identify facilitators and barriers to collecting PROs. The goals of this paper are to: (1) report the results of this survey, (2) present current performance on PRO data

Abbreviations

CHAQ, child health assessment questionnaire; cJADAS, clinical juvenile arthritis disease activity score; EMR, electronic medical record; JIA, juvenile idiopathic arthritis; LHN, learning health network; OMERACT, outcome measures in rheumatology; PR-COIN, pediatric rheumatology care and outcomes improvement network PR-COIN; PRO, patient-reported outcome; PROM, patient-reported outcome measure; PROMIS, patient-reported outcomes measurement information system.

reporting in the PR-COIN registry and (3) present results of interviews highlighting sites that successfully implement systems to collect and transfer completed PRO data to the electronic medical records (EMR) and registry.

Materials and methods

The PR-COIN registry was approved by Seattle Children's Institutional Review Board (IRB), which serves as the IRB of record for Seattle Children's Hospital.

A REDCap survey was sent to the lead representative for each PR-COIN site. Lead representatives were asked to consult with their site members prior to completing the survey. Survey questions included how PROs were collected, which PROs were collected, facilitators and barriers to collection (Supplementary 1). Quantitative data were analyzed using descriptive statistics and qualitative data were thematically analyzed.

Registry data was analyzed to better understand the completion rates of patient reported data such as morning stiffness, pain intensity scores, physical function, and overall well-being.

The interviews with site physician leaders were deemed exempt by Hospital for Special Surgery's Institutional Review Board.

PR-COIN conducts biannual meetings where sites within the LHN share experiences to facilitate learning. Four physician leaders who previously reported successful implementation of PRO collection were invited to participate in a one-time virtual interview (with NP) where they shared how PROs were collected at their site, identified key resources that facilitated documentation of PROs in their EMR, barriers they had to overcome, and share best practices (Supplementary 2). A summary of the interview was provided to each participant for review and approval.

One-on-one interviews of physician leaders were conducted by videoconference by NP and audiotranscribed. Preliminary thematic analysis was conducted independently by NP and EMM, and agreement on major themes achieved through discussion. Subsequently, two separate reviewers (IG and SJ) identified themes using inductive thematic analysis utilizing NVivo 15 software by Lumivero (7).

Results

All 23 PR-COIN sites responded to the survey. PROs are collected by 21/23 (91%) sites which variably measured patients' perception of their condition or symptoms of their condition, self-management, medication side effects, ability to do activities of daily living, and mental health status (Table 1). PROs were collected for both clinical and/or research purposes. The top three collected PROs for both clinical and research purpose were arthritis-related pain intensity score, morning stiffness and physical function as measured by the Childhood Health Assessment Questionnaire (CHAQ) score (8). The PR-COIN registry collects morning stiffness using delineated increments of time and the survey responses reflect the number of sites reporting morning stiffness in the registry. Five of 23 (21.7%) sites indicated that their institution had mandated certain questionnaires be collected throughout their institution e.g., assessments of mental health and suicide screening.

TABLE 1 Patient reported outcome measure collection in PR-COIN.

Patient reported outcome measure		
Pain-intensity score	19/23	82.6%
Morning stiffness	18/23	78.3%
Child health assessment questionnaire (CHAQ)/Health assessment questionnaire (HAQ)	17/23	73.9%
Patient global-overall well-being	14/23	60.9%
Patient global assessment	13/23	56.5%
Review of systems	13/23	56.5%
PROMIS-Pain interference	6/23	26.1%
Transition readiness	6/23	26.1%
Patient health questionnaire-9 (PHQ-9)	5/23	21.7%
PROMIS-upper extremity	5/23	21.7%
Patient global-disease activity	4/23	17.4%
PedsQL RHE child	4/23	17.4%
PedsQL RHE parent	4/23	17.4%
PROMIS-mobility	4/23	17.4%
PedsQL core child	3/23	13.0%
PedsQL core parent	3/23	13.0%
Bath ankylosing spondylitis disease activity index (BASDAI)	2/23	8.7%
Patient health questionnaire-2 (PHQ-2)	2/23	8.7%
PROMIS-depressive symptoms	2/23	8.7%
Juvenile arthritis functional assessment report (JAFAR)	1/23	4.3%
Methotrexate intolerance severity score (MISS)	1/23	4.3%
PROMIS-anxiety	1/23	4.3%
PROMIS-fatigue	1/23	4.3%
Quality of my life (QoML)	1/23	4.3%
EQ-5D	0/23	0.0%
Juvenile arthritis functional status index (JASI)	0/23	0.0%
Juvenile arthritis functionality scale (JAFS)	0/23	0.0%
Juvenile arthritis multidimensional assessment report (JAMAR)	0/23	0.0%
Juvenile arthritis quality of life questionnaire (JAQQ)	0/23	0.0%
Outcome measure child health questionnaire (CHQ)	0/23	0.0%
Pain symptom assessment tool (PSAT)	0/23	0.0%

Interestingly, the patient global assessment of overall well-being was collected more often for research purposes rather than clinical purposes. The patient global assessment score was collected on varying scales (0–10 vs. 0–100 range) and varying increments (1 vs. 0.5 vs. 0.1 unit). Nine of 23 (39%) sites indicated that they planned to add additional questions or questionnaires to measure PROs, or PROMs such as PROMIS short form measures (9) in the future.

One site reported not distributing questionnaires to their patients to complete, whereas seven sites indicated that 100% of their patients received PROMs (Figure 1). Eleven sites reported high reliability of completion of the distributed PROMs, with a 76%–99% completion rate (Figure 2).

Respondents reported that PROMs were collected using both paper and/or electronic methods, with many sites 11/23 (47.8%) collecting PROMs only on paper, fewer 7/23 (30.4%) collected PROMs only digitally, and 5/23 (21.7%) collected PROMs both on paper and digitally. When collected digitally, sites indicated that PROMs were administered using a variety of methodologies:

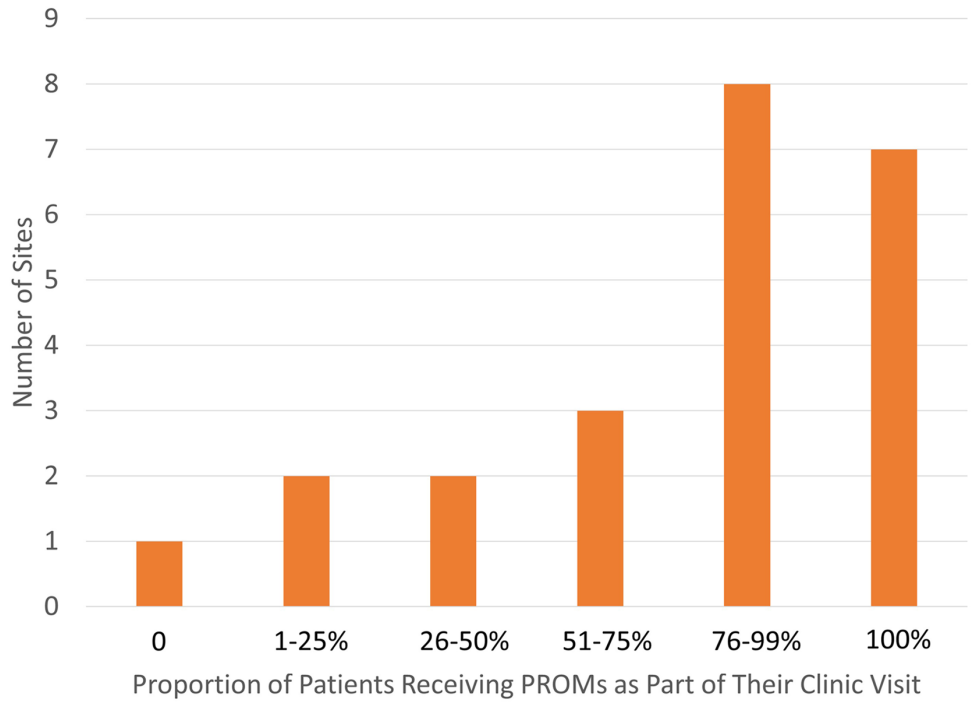

FIGURE 1
Patients receiving PROMs at PR-COIN sites.

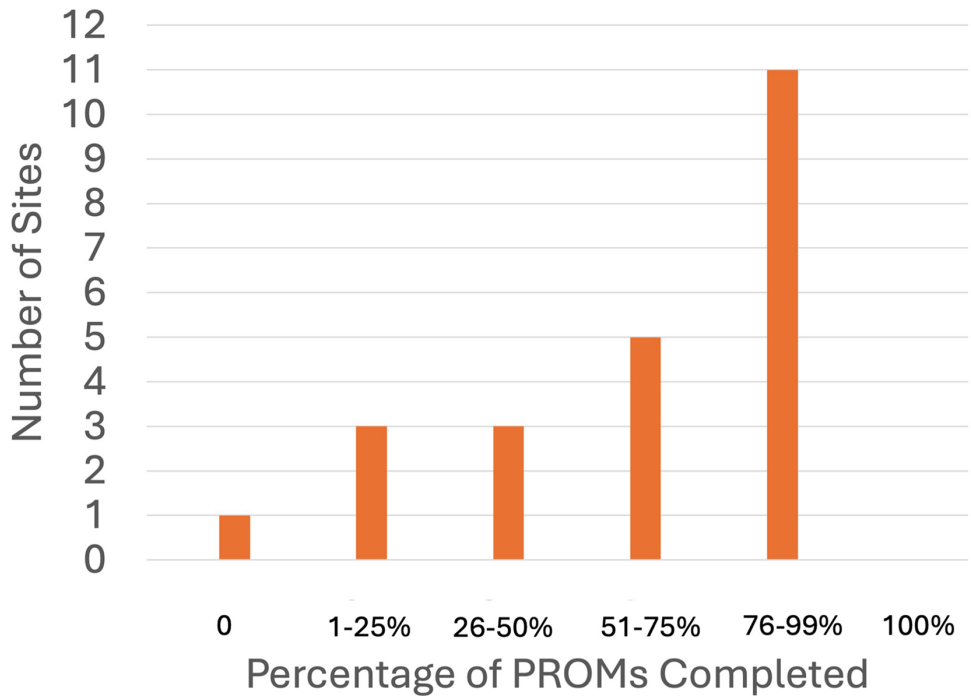

FIGURE 2
PROMs completion at PR-COIN sites.

a tablet with data flowing directly into the EMR or REDCap, via patient portal into the EMR, and website.

All sites used EMR systems to document their patient encounters 17/23 (73.9%) Epic (10) 3/23 (13%) Cerner (11), and AllScripts (12) 3/23 (13%).

Sites reported involvement of a variety of individuals in the administration of PROMs, including physicians, nurses, medical assistants, front desk staff, research team members, volunteers, and self-administration by patients/proxies. More than 75% of respondents reported patients had the ability to self-administer PROMs via a tablet in clinic or patient portal survey. PROs were usually completed by either patient or proxy before or during the appointment. Most sites reported that PROMs were completed by more than half of the patients (Figure 1).

Respondents estimated that staff spent three minutes administering PROMs compared to six minutes for patients and proxies to self-administer.

The majority of sites (14/23, 60.9%) reported that PROM scores were manually calculated. Five of 23 sites (21.7%) indicated that the score was electronically calculated, whereas 4 of 23 (17.4%) used either electronic or manual methods to calculate the score. In cases where the scores were manually calculated, individuals performing the calculations were: physician, practitioner, nurse, trainees, medical assistant, research team member, and volunteer.

Respondents estimated that clinical or research staff took about four minutes to manually enter scores into the EMR. Although some sites reported their patients and proxies could complete their PROMs electronically, 5/12 (42%) sites reported that scores could not automatically be imported into the patient's EMR.

Respondents indicated that availability of personnel, training and culture were the greatest facilitators, whereas limited time and lack of staff were the greatest barriers to PROM completion.

Self-reported completion of PROs in this survey cannot be extrapolated to completeness of data entry in the PR-COIN Registry. Data from the PR-COIN registry showed that across the 14 centers submitting PROs, morning stiffness was collected at all sites. Arthritis-related pain scores were collected from 13 of 14 sites (92%) and patient global assessment of overall well-being scores from 13/14 sites (92%). Measures of physical function included the CHAQ, Patient-Reported Outcomes Measurement Information System (PROMIS) mobility and PROMIS upper extremity measures. Ten of 14 centers (71.4%) completed at least one of these three measures for physical function while only two sites completed all three measures. The range of completion was 93%–98% for overall well-being and pain intensity scores and 69%–94% for physical function for the top four performing centers reporting data.

Case studies

Four physician leaders in PR-COIN who previously reported successful collection of PROs were interviewed and shared their operational processes and lessons learned.

PRO collection

Each of the high-performing sites initially started collection of PROs on paper. Prior to joining the LHN, PROs were collected primarily for the purposes of research registries or clinical trials, rather than for clinical care of the patient. Two of these sites had a strong culture of collecting PROs and reliable processes of paper-based data collection preceding joining PR-COIN. One site linked the collection of "Review of Systems" items to collection of pain score and overall wellbeing which may have contributed to the high collection rate. One site noted that a prior workflow utilized REDCap as the only access to PROs, which hindered physician engagement with the data. Physician interaction and access to data subsequently improved after the incorporation of an automated form in the EMR, which eliminated the need to log into a different system.

Barriers

Time limitations were often cited as a barrier to collecting, reviewing, and acting on PROs results at point-of-care. A perceived or actual increased workload is a major barrier to collection and utilization of PROs by the clinical team.

All sites reported that utilizing the clinical Juvenile Arthritis Disease Activity Score (cJADAS) (13) in "treat to target" (5) discussions was the most common scenario where PROs are used by their colleagues. However, not all physicians at each site necessarily discussed answers of the PROs with patients, primarily citing time constraints as a barrier. Physicians were also concerned that additional time would be needed to better understand any discrepancies between the patient global assessment of overall wellbeing and the physician global assessment of disease activity. In general, this has not discouraged efforts for PRO collection across the rheumatology teams and, as a group, physicians recognized the importance of collecting patient perspectives.

While increased workload for physicians was cited as a common barrier to administration of PROs, use of automated systems to offset the workload was identified as a critical facilitator for PRO collection by each of these sites.

Other barriers to collection of specific PROMs include obtaining permission/license to use certain surveys (Table 2).

Facilitators

Two of the most effective facilitators of successful collection of PROs identified among all four sites were the roles of an engaged leadership team and information systems team for initial implementation and ongoing maintenance. An established culture of collecting PROs was also cited as an important facilitator (Table 2).

Leadership engagement was identified as a strong facilitator at all the sites, with three sites citing specific examples. A unique facilitator, conceived by the quality improvement physician champion and the section chief, employed at one site is a

TABLE 2 Self-identified facilitators of PROs collection rates in PR-COIN.

Facilitators to PRO collection
PRO integration to EMR and learning network registry
Minimal burden (e.g., time and effort) for physician
Patient engagement in selection of measures
Presence of a clinical champion and project manager to encourage adoption by clinical team
Physician review of and use of PRO responses with patients to track patient's health status
Departmental leadership support and resources
Previous experience collecting PROs
Fostering a culture of PRO collection as standard practice
Adequate staffing to assist with collecting and documenting PROs
Adequate training for staff and clinicians
Presence of discrete response options
Automated reminders to both patients and staff members to complete/collect PROs
Barriers to PRO collection
Limited time
Lack of staff and resources administer PROs and enter data
Lack of resources to build IT infrastructure and oversee data transfer
Additional workload which interrupts clinical workflow
Lack of buy-in from individuals with interests (physicians, institutional leadership)
Low priority for institution
Lack of understanding of importance of PROs from patients/proxies
Lack of or suboptimal automation of PRO collection
Lack of adequate training of staff in data collection and data validation
Concern by physicians about alignment of PROs scores with physician assessment
Lack of availability of PROMs in different languages
Lack of interface to share and discuss PROs with patients
Difficulty standardizing PRO responses

PRO, patient reported outcomes.

physician incentive, linked to the documentation of physician global score and joint count in a prespecified proportion of all patient encounters. While these are not PROs, the two measures coupled with a measure of patient overall well-being comprise the cJADAS (13). Providing maintenance of certification is another motivator for physician participation at this site.

Automated collection and calculation of PRO scores were consistently identified as a facilitator of successful PRO collection and clinician engagement. One site leader said, "the biggest thing I would say is to get the questionnaires electronic." Another leader agreed, stating because PRO collection is "fully automated on the tablet, the burden for the nurses is really minimal." One center described that the automated calculation of a composite disease activity measure, cJADAS (13), which incorporates a PRO ("overall well-being"), as a motivation for the clinical team to ensure that patients complete the PROM. The reason providers were invested in the PROM completion is because the cJADAS is the basis of a "treat to target" intervention used by providers to improve patient outcomes (5) and part of the critical data set of PR-COIN. Three of four sites indicated that the cJADAS is automatically calculated by the EMR. Electronic data transfer to the PR-COIN registry platform (4), is automated at three of the four sites interviewed. The fourth site currently relies on the nursing staff to screen for eligible patients prior to the office visit and to ask PRO questions during the rooming process. Scores are manually calculated by the physicians. This site plans to implement an automatic calculation model in the future. Successful transfer of data to the registry at this site was attributed to having a dedicated staff member who manually uploaded data. Thus, the involvement of an information systems team to build direct electronic transfer and provide tablets was identified as an important facilitator that would remove the burden of PRO collection from the clinical and research staff.

Physician engagement was another important facilitator. One site expressed the importance of having a project manager and physician champion with experience with quality improvement methodology, identification of clinically meaningful measures and practical aspects of implementation, such as frequency of releasing surveys.

Buy-in from other clinical staff was also cited as important. At one site, nursing staff were integral to the PRO collection process. There was a high level of engagement from nursing leadership (where they had the same nurse leader for the past 10 years).

Patient engagement is critical to the successful collection of PROs. One site specifically involved patients in designing the surveys, with careful attention to minimize burden and include PROs that patients felt were important. All sites noted that physician acknowledgment and utilization of PROs motivated patients to complete surveys. It provides "positive reinforcement that we are listening to them." Other sites agreed, citing culture where "patients are just used to filling these out", generally resulted in high fidelity, as did patient portal access and short length of surveys. One site reported conducting a study on the utilization of open note access, where patients were encouraged to review their own office notes. Patients reported that their efforts were validated when they saw their responses incorporated into their physician's notes.

Lessons learned

Other important components of successful implementation included: practicing patience, making small, incremental changes, and establishing a unified workflow with the entire clinical team prior to implementation in order to maximize engagement.

To facilitate physician interaction with PROs at point of care, data were made available through multiple methods. Each site had a distinct tab in their EMR where PROs can be viewed. They also had a note template, which "pulled in" patient answers, making them immediately visible to the treating physician. Several sites also presented data in a flowsheet or dashboard, allowing results to be tracked over time, facilitating discussions with patients during their visit. One site programmed PROs to be released at regular intervals, with the option of setting up best practice alerts at pre-specified intervals (e.g., three months after treatment change) to review "treat to target" goals.

Training on the workflow for collecting PROs and their use in direct patient care, as well as the introduction to quality improvement efforts were provided to all new trainees and faculty at each of these sites.

Regular meetings led by the physician champion with the clinical team to review data and disease activity scores were deemed important at reinforcing the collection of PROs and

maintain high rates of completion. Sharing the impact of improvement efforts on a regular basis "swayed even the skeptics in the group." At one site, graphs of anonymized PRO completion rates for the division were published monthly and higher-performing providers were invited to share their best practices with the team. Additionally, the physician champion had conducted annual one-on-one meetings with each clinician to review individual's and center performance (completion rates and overall disease scores/remission) and provided support to improve rates.

Participation in PR-COIN was credited for providing structure, motivation, and justification for existing PRO collection workflows. At one site, the formalized collection of PROs was established after joining PR-COIN. The physician leader at this site noted that the contributing data to a registry validates the value of effort from the physicians, nurses and data team.

In addition to technical support and guidance from PR-COIN, these leaders proposed the creation of a formal guide to improve PRO collection, establishing high-level steps and milestones, performance objectives of division chief, quality improvement physician champion, information systems teams, parent engagement, and ancillary and research staff. Furthermore, formal recommendations from an LHN can serve as a powerful advocate to persuade local hospital leadership of the significance and impact of PRO collection.

All four sites envisioned the creation of a patient-facing platform in the future, which would enable patients to view their PROs over time. Three sites also planned to have PROs available in other languages in order to improve delivery of care and communication with non-English speaking patients.

Discussion

As healthcare increasingly focuses on patient-centered care, it is imperative that healthcare providers, researchers, and policy makers collectively support and adopt processes to enable reliable and complete collection of PROs. Reaching a consensus on a core set of PROs that accurately reflect patients' needs and desires, while minimizing the burden on patients and proxy reporters, is a primary step to achieving this goal. This would serve as a foundation for standardizing processes and systems to optimize the collection and use of PROs to improve health outcomes.

Through a consensus-based approach with patients, parents/caregivers and healthcare providers, the Outcome Measures in Rheumatology (OMERACT) Juvenile Idiopathic Arthritis (JIA) Workgroup has created a detailed definition and description for the two target domains in the patient perception of overall well-being related to disease (6, 14). Through the PR-COIN Parent Workgroup, patient and parent voices have informed the PROs collection (3).

Our survey revealed varying PRO collection rates and process across the LHN. The self-reported nature of the survey has limitations as it may not accurately reflect the completion of PRO data fields or reliable transfer into the shared LHN registry. Integration of PROs into EMRs was identified as a facilitator to PRO collection from both the survey and interviews we conducted. EMR integration requires an upfront investment but will enable healthcare providers to efficiently collect data longitudinally. As an LHN, building a system that digitally collects PROs, having alignment of PROs across the network and the collection of responses using standardized scales would facilitate the network's ability to compare outcomes of treatment across network sites using PROs.

To increase the incorporation of patients' perspective in their clinical care, an equally critical consideration is accessibility of PROs in the EMR for both clinicians and patients. Involving clinicians in the design of PRO displays may improve their ability to act on the information while minimizing burden of additional "clicks". Our interviews revealed that when patients see their data being utilized in treatment decisions, it helps them understand the rationale for completing and motivates them to complete PROs. Accessibility of PROs for clinicians and patients highlights the importance of technical support to build and maintain an accessible interface.

In addition to the importance of integrating PRO data into the EMR and ensuring accessibility of PROs for both clinicians and patients, the survey and interviews revealed other facilitators for successful PRO collection: minimizing clinician time and effort (i.e., in administration/calculation), having a designated physician champion and project manager, providing feedback on collection rates to the clinical team, and fostering a culture that values PRO collection within the department or institution. Notably, each of the four PR-COIN sites interviewed had an established paper-based PRO collection process before integrating it into the EMR. This experience likely assisted in establishing the feasibility of PRO collection and utilization prior to hardwiring this process electronically. While there is no single effective model, considerations for planning and implementation are central to successful and sustainable PRO collection.

Lessons learned from other successful organization-wide implementation of PRO collection and utilization (15, 16) echo those learned from our LHN. Key factors include physician and administration engagement, presence of a clinical champion, prior experience with PRO collection, and payer incentive contracts.

Based on these insights, PR-COIN is now well-positioned to develop a toolkit (17). The toolkit will outline sample workflows, implementation strategies and other resources for collecting PROs, similar to the approach which has been used in a learning health network for rheumatoid arthritis patients (18).

For a LHN focused on patient outcomes, it is essential to administer and complete PROs with high reliability to measure performance and guide improvement in areas prioritized by patients. Currently, there is limited guidance on standardization of PRO collection within a LHN, resulting in variable rates of completion. Through understanding our current ability to systematically collect PROs across all PR-COIN sites and exploring successful implementation of PRO collection both within and outside our network, we share lessons learned and identify key factors that contribute to the successful spread of this important practice.

Ethical considerations

The PR-COIN registry was approved by Seattle Children's Institutional Review Board (IRB), which serves as the IRB of record for Seattle Children's Hospital for following relying participating sites: Stanford University, University of Mississippi, Children's Wisconsin, Northwell Health/Cohen Children's Medical Center, Baylor College of Medicine/Texas Children's Hospital, University of Minnesota, Phoenix Children's Hospital, Nationwide Children's Hospital, Medical University of South Carolina, Hospital for Special Surgery, Hackensack Meridian Health, Cincinnati Children's Hospital Medical Center, Children's Mercy Kansas City, Children's Hospital of Philadelphia, Boston Children's Hospital, and University of Alabama at Birmingham. Due to institutional regulatory policies and local or provincial laws and regulations. The registry was approved by a local IRB for 6 participating sites: Levine Children's/Atrium Health (Charlotte, NC, United States), London Health Sciences Centre/Lawson Health Research Institute (London, ON, Canada), McMaster University (Hamilton, ON, Canada), Nemours Orlando (Orlando, FL, United States), Penn State Children's Hospital (Hershey, PA, United States), and The Hospital for Sick Children/SickKids (Toronto, ON, Canada).

PR-COIN uses a collaborative learning health system approach to improve quality of care and outcomes for children with JIA. PR-COIN currently has 23 participating sites from academic pediatric medical centers throughout the United States and Canada. PR-COIN is led by a coordinating center which provides quality improvement consultation, quality improvement education, maintenance of certification opportunities, data management, data analytics, legal and regulatory supervision, project development and oversight, and overall support to the network.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The requirement of ethical approval was waived by Hospital for Special Surgery Institutional Review Board Seattle Children's Institutional Review Board for the studies involving humans because IRB exemption obtained for research that only includes interactions involving interview procedures, if at least one of the following criteria is met: (ii) Any disclosure of the human subjects' responses outside the research would not reasonably place the subjects at risk of criminal or civil liability or be damaging to the subjects' financial standing, employability, educational advancement, or reputation. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation was not required from the participants or the participants' legal guardians/next of kin because IRB exemption for reason any disclosure of the human subjects' responses outside the research would not reasonably place the subjects at risk of criminal or civil liability or be damaging to the subjects' financial standing, employability, educational advancement, or reputation.

Author contributions

NP: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. EM: Conceptualization, Methodology, Project administration, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing. MR: Writing – review & editing. BG: Writing – review & editing. JH: Writing – review & editing. TL: Writing – review & editing. YG: Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Project administration, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fped.2024.1443426/full#supplementary-material

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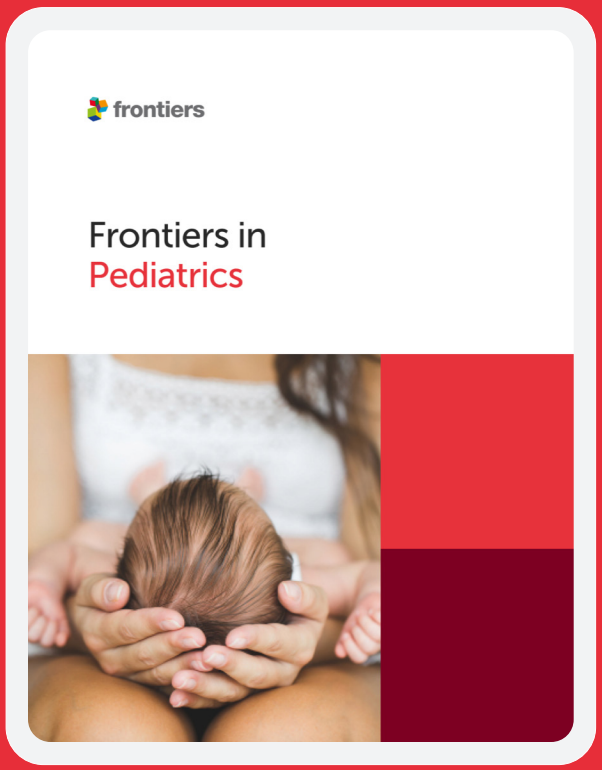

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