Juvenile Idiopathic Arthritis

Study protocol - April 2023

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Introduction

Juvenile idiopathic arthritis (JIA) is a term that covers a heterogeneous group of conditions characterised by inflammatory arthritis with onset before the age of 16 years and lasting for more than 6 weeks.[1] The causes of JIA are poorly understood but likely relate to genetic and/or environmental factors.[2]

JIA is an 'umbrella' term which covers different sub-types including:

- Oligo-articular JIA
- Extended Oligo-articular JIA
- Poly-articular JIA (RF –ve)
- Poly-articular JIA (RF +ve)
- Systemic-onset JIA
- Psoriatic JIA
- Enthesitis-related Arthritis

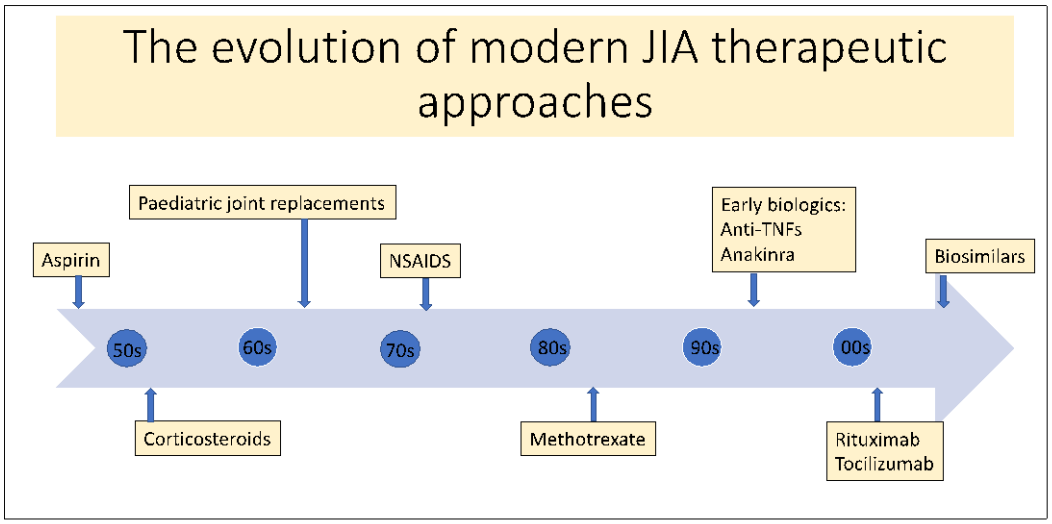
JIA can also affect other parts of the body, most commonly the eye, with between 10% to 20% of children developing JIA associated uveitis (potentially blinding inflammation in the eyes).[3]

The incidence and prevalence of JIA are poorly understood. In 1996, Symmons et al published data from the British Paediatric Rheumatology Group National Diagnostics Register estimating an incidence of around 1:10,000 children with an overall prevalence in childhood of 1:1000 .[4] A more recent study used primary care electronic health record data to calculate contemporaneous incidence and prevalence data. The age-standardised incidence rate was 5.61:100,000 population with an age-standardized prevalence rate of 43.5:100,000.[5] A relatively recent literature review used 43 JIA epidemiology articles and 2010 European age and gender pooled rates to estimate the number of cases across Europe, reporting a direct standardised incidence rate of 8.2 ([7.5–9.0]/100,000) and prevalence of 70.2 ([62.9–78.1]/100,000).[6] Authors commented on a significant between-study variation in incidence and prevalence data. This may reflect a true variation associated with ethnicity and/or geography but JIA is a relatively rare group of diseases, and few countries have well-established systems collecting complete incidence and prevalence data. Low awareness of the condition, and changes in terminology, culminating in the development of the International League of Associations for Rheumatology (ILAR) classification criteria[7] are likely to further complicate the reporting of JIA cases.

In 2020/21, in England there were 11,875 hospital admissions in young people aged 0-24 with a diagnosis code for juvenile rheumatoid arthritis (M08). Within this group, 9,207 admissions were for young people aged ≤17 years, and a majority were on a day-case basis.[8] The term 'juvenile' refers to the age at onset and historical data suggest that around half of children with JIA continue to have episodes of active arthritis as adults.[9]

Standard treatment for JIA includes one or more of non-steroidal anti-inflammatory drugs (NSAIDS) for analgesia, intra-articular corticosteroid injections, systemic corticosteroids, methotrexate and/or other disease modifying anti rheumatic drugs (DMARDs) such as sulfasalazine, and/or modern biologic or small molecule therapies.[10, 11, 12] Children and young people are treated in accordance with NICE guidance[12, 13] and/or agreed NHS England treatment pathways.[14] Access to a multidisciplinary team with expertise in paediatric rheumatology is essential if children with JIA are to achieve disease control. Children should be managed within established clinical networks linked to tertiary paediatric rheumatology units.[15, 16]

The evolution of modern JIA therapeutic approaches



JIA is a diagnosis of exclusion which can mean that a "high index of suspicion for other differential diagnoses is necessary"[17] particularly when toddlers and children with "limited expressive skills" develop symptoms.[18] Variation in presentation can frequently lead to misdiagnosis in the early stages of disease and/or unnecessary investigation and treatments prior to the actual diagnosis being revealed.

Additionally, the insidious nature of the onset of symptoms can lead to delay in treatment and referral to specialist centres. Variation in organisation of specialist services within Trusts/Health Boards can complicate the pathway further, again leading to a delay in recognition of disease as well as referral to specialist services. Indeed, the British Society for Rheumatology (BSR) workforce report indicated that there is a significant lack of exposure to rheumatology in formative AHP, pharmacist and medical training.[19]

Multicentre observational cohort study datasets clearly demonstrate that a significant proportion of children and young people (CYP) with JIA struggle to achieve inactive disease during the first two years of disease.[20, 21, 22] Achievement of inactive disease is associated with disease subtype and is less likely in the presence of diagnostic delay,[23, 24] suggesting that improvements in the quality and consistency of clinical care have the potential to significantly impact early clinical outcomes. Diagnostic delay is a real concern in the UK where presentation to specialist rheumatology services may be via troublingly circuitous routes. A lack of awareness among GPs can further add to these delays.[25]. Improving early clinical outcomes is an important way to prevent undesirable long-term consequences of disease, relating to prolonged treatment with immunosuppressive medication and/or repeated surgical interventions. A high proportion of patients with JIA will continue to have joint pain and/or disability in adulthood – either related to ongoing episodes of active arthritis or irreversible joint or eye damage.[26, 27]

In response to growing concerns about delay in access to care and inequity of clinical services, the British Society for Paediatric and Adolescent Rheumatology (BSPAR) and the Arthritis and Musculoskeletal Alliance (ARMA) led the development of Standards of Care (SOC) for CYP with JIA between 2007 and 2010.[16, 28] Key philosophies underpinning the SOC included empowerment of patients and carers in treatment plans and a holistic approach to the provision of care. The SOC became acknowledged internationally as minimum standards for clinical services,[29] underpinning a steadily increasing awareness of the impact of delay in

access to care.[20] A 2013 multi-site UK audit against key SOC demonstrated considerable variation in service delivery and time to access specialist care.[30]

Since 2013, the UK Paediatric Rheumatology community has worked closely with the BSR towards improved JIA data collection. UK-wide collaboration with CYP, families, clinical teams and academics has resulted in development and piloting of the CAPTURE JIA dataset, [31] highlighting the importance of paediatric data collection and the increasingly clear need for significant and ongoing investment to enable effective data collection and analysis. BSR and the GIRFT team have funded a new JIA learning collaborative (JIA Learn), with the aim of improving patient reported experiences and outcomes of care for CYP with JIA by January 2024.[32, 33] Unfortunately, effective national level CAPTURE-JIA would require significant and ongoing investment and the relative rarity of JIA renders this level of investment difficult to secure.

In summary, there is still much room for improvement to the quality of care available to CYP with JIA. Improved quality of care would reduce the impact of this long-term condition on physical and emotional health, quality of life and access to education/work, and inequalities. A key precursor to implementation of meaningful improvements in the quality of care is access to national level data relating to disease prevalence and patterns of recognition, referral, diagnosis and treatment.

Guidelines and standards

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Aim and objectives

Overall aim:

To review the quality of care in children and young adults (0-24 years) with Juvenile Idiopathic Arthritis (JIA).

Objectives

Organisational

To review:

- The referral process (including GP to secondary care, secondary care to tertiary care, and the availability of rapid access clinics)
- Networks of care (including shared care)
- Protocols for the management of JIA
- Treatments (including variation in treatment choice (depending on practitioners))
- Multidisciplinary team working (including members of the MDT)
- Access to equipment
- Job planning and how local services are managed (including staff:patient ratios)
- The availability of peer support
- Transition (including adherence to NICE guidelines)
- Education, training and research
- Audit (including the use of PROM/PREM)
- Variability in access to care

Clinical

To review:

- Recognition of disease (patterns of presentation prior to diagnosis, under recognition, misdiagnosis and overdiagnosis)
- Pathways of care (including primary, secondary and tertiary care, sole tertiary care treatment, and shared care)
- Investigations for the initial diagnosis (including access to imaging)
- Appropriate and timeliness of first assessment and treatments (by rheumatology and ophthalmology)
- Multidisciplinary team approach and access to the MDT
- Specialist nurse involvement and role
- Type of treatment – NSAIDs, steroids (local, oral, IV, IA), DMARDs, Biologics, JAK inhibitors (including choice of treatment and why, timing of treatment, availability of treatment, monitoring of treatment and information available to young adults and parent carers)
- On-going assessment of disease activity using clinician derived outcome measures and PROM/PREM
- Follow-up arrangements
- Transition
- Variability in clinical care dependent on type of care (i.e. shared care etc)
- Discrimination (including delays in diagnosis and in education)

Methods

Early young person and parent carer involvement

As part of the early scoping of this study, we have undertaken a series of focus groups with young adults and parent carers to identify the areas of care important to review, and to

ensure a patient-centred study. The results of these have fed into the development of the study aims and objectives, and a summary is included in Appendix 1.

Inclusion criteria
Children and young adults aged 0 to 24 years, inclusive, with JIA.

Children and young adults will be identified for inclusion in two ways:

A nominated study contact will be asked to identify a sample of children and young adults with JIA under their organisation's rheumatology service over a two-year period from the 1st April 2021 to 31st March 2023 (method detailed below in case identification) and populate the Patient Identification Spreadsheet with their details. This will enable us to identify a sample of children and young adults receiving care via the outpatient department.

In addition to this, local reporters will be asked to identify children and young adults who presented to their organisation (including day cases and outpatient attendances where available) over the same two-year period using the following ICD10 codes. Corresponding Snomed codes will be made available where required.

ICD10 codes for inclusion

- L40.54 Psoriatic juvenile arthropathy (where available)
- M08.0 Juvenile rheumatoid arthritis
- M08.1 Juvenile ankylosing spondylitis
- M08.2 Juvenile arthritis with systemic onset
- M08.3 Juvenile polyarthritis (seronegative)
- M08.4 Pauciarticular juvenile arthritis
- M08.8 Other juvenile arthritis
- M08.9 Juvenile arthritis, unspecified
- M09.0 Juvenile arthritis in psoriasis
- M09.8 Juvenile arthritis in other diseases classified elsewhere

Exclusions

- M09.1 Juvenile arthritis in Crohn disease [regional enteritis]
- M09.2 Juvenile arthritis in ulcerative colitis

Data sampling timeframe
The timeframe from which data will be sampled will be the 1st April 2021 – 31st March 2023.

Participating providers of healthcare
All providers of healthcare across England, Wales and Northern Ireland, where children and young adults with JIA might be cared for will be asked to participate in the study. This will include acute, community and independent organisations. Primary care practices will be asked to participate if it is identified that a patient selected for inclusion in the study was under the care of their practice.

Incidence and prevalence of the exemplar conditions
Table 1. Nationally collated hospital admission data
Hospital Episodes Statistics (HES) 2021/22; Patient Episode Database for Wales (PEDW) 2020/21.

	HES data 2020/21 (Age 1-24 years)	PEDW data 2020/21
Diagnosis (ICD10) code		
Arthropathic psoriasis (L405)	865	13
Juvenile rheumatoid arthritis (M080)	1,977	18
Juvenile arthritis with systemic onset(M082)	875	12
Juvenile polyarthritis (seronegative) (M083)	2,239	4
Pauciarticular juvenile arthritis (M084)	1,674	3
Other juvenile arthritis(M088)	1,529	8
Juvenile arthritis, unspecified (M089)	3,581	114
Juvenile arthritis in psoriasis (M090)	418	0
Juvenile arthritis in other diseases classified elsewhere (M098)	43	0

Study promotion
Prior to data collection, NCEPOD will contact all healthcare organisations providing care to this group of children and young adults. The study will be promoted to children and young adults, and parent/carers via patient groups, third sector organisations, NCEPOD Local Reporters (sending the study poster on to the relevant departments), via any study contacts recruited, and via the relevant Colleges and Associations.

Study method test
The data collection methods and data collection tools will be tested to ensure they are robust before the full study is run.

Methods of data collection

There will be five main ways of collecting data for the study:

1. Young person/parent carer views will be collected through focus groups and an online anonymous survey. We will work with Local Reporters, and relevant charities (e.g. Versus Arthritis, The National Rheumatoid Arthritis Society, Juvenile Arthritis Research) to encourage involvement.

2. Clinician views will be collected through an online anonymous survey. We will work with Local Reporters and study contacts to encourage involvement from clinicians and commissioners.

3. An organisational questionnaire will be sent to all providers of healthcare where children and young adults with JIA might be cared for.

4. Clinical data collection: For a sample of children and young adults diagnosed with JIA between 1st April 2019 – 31st March 2023, a questionnaire will be sent to all clinicians responsible for providing ongoing rheumatology care. Questionnaires will be sent to clinicians working in primary, acute, community and independent organisations.

5. Case note review: Copies of selected extracts of case notes will be collected for peer review.

Further details on the methods of each method of data collection are given below.

1. Anonymous online young person and parent carer survey and focus group interviews
The survey and focus group interviews will gather data on the young person and parent carer views of the services available to them. The data will not be linked to any other aspects of data collection.

2. Anonymous online clinician survey
The survey will gather data on clinician views of the services available for them to provide to children and young adults with JIA. The data will not be linked to any other aspects of data collection.

3. Organisational questionnaire
Data collected will include information around the referral process, networks of care, the use of protocols for the management of JIA, treatments, multidisciplinary team working, access to equipment, job planning, transition, and audit. Questionnaires will be sent to all organisations participating in the study via the online questionnaire system.

4. Clinical data collection
Identification of children and young adults for inclusion
Two methods will be used to identify children and young adults to include in the study:

a) At the start of the study the Local Reporter will be asked to set up a study contact within the rheumatology department (e.g., the rheumatology lead). The study contact will be asked to identify a sample of children and young adults under the care of the rheumatology service between 1st April 2021 – 31st March 2023, from both paediatric and adult services. This will enable us to identify a sample of patients from the outpatient community. Where these details are recorded electronically this should be straightforward. Where the details are not recorded electronically, the study contact will be asked to contact NCEPOD.

The study contact will be asked to complete the patient identification spreadsheet with the details of the sample of children and young adults under the care of the rheumatology service during the data sampling time frame. The data fields requested will include NHS number, hospital number, date of birth, sex, postcode, arthritis diagnosis, date of diagnosis (mm/yyyy) or the date of first appointment with a rheumatologist or a general paediatrician with a special interest in rheumatology. We will also request the details of the primary care practice name and the details of any other organisations providing arthritis care, so we are able to track healthcare across organisations.

b) The Local Reporter will also be asked to identify children and young adults who presented to their organisation between 1st April 2021 – 31st March 2023 using the included ICD10 codes. This will include day case admissions and outpatient attendances where appropriate.

The Local Reporter will be asked to complete the patient identification spreadsheet with the details of all children and young adults who received care at their organisation during the data sampling time frame. The data fields requested will include NHS number, hospital number, date of birth, sex, postcode, date of diagnosis (mm/yyyy) (where available), date of most recent admission to this hospital, included ICD10 codes (primary and all), OPCS codes, the date of first encounter to this organisation with an included ICD10 code; date of first appointment with a rheumatologist or a general paediatrician with a special interest in rheumatology; number of outpatient appointments attended; details of the lead rheumatology clinician. We will also request the details of the primary care practice name

and the details of any other organisations providing arthritis care, so we are able to track healthcare across organisations.

Tracking healthcare across multiple organisations
The NHS number and date of birth will be used to track healthcare across multiple organisations. The details of providers of care outside the organisation submitting the data will be requested on the patient identification spreadsheet, and in the clinician questionnaires. If a child or young adult is identified as receiving care from an organisation, but was not included on their initial data spreadsheet, the local reporter of that organisation will be contacted, before questionnaires are sent out, and asked to confirm whether they are known to them based on the available NHS number.

Clinician questionnaires
Two questionnaires will be used to collect clinical data for this study:

1) Clinician questionnaire
2) Primary care combined clinician and organisational questionnaire

Clinician questionnaire
Clinician questionnaires will be sent for a sample of children and young adults, where the diagnosis was made between the 1st April 2019 – 31st March 2023. The clinician questionnaires will be sent to the NCEPOD Local Reporter for dissemination via the online questionnaire system. A reminder will be sent at six weeks and ten weeks where the data is outstanding. Up to 15 children or young adults per hospital will be sampled for inclusion in the study. In addition to this, a questionnaire will also be sent if the hospital is identified as providing care to a child or young adult identified for inclusion in the study via another organisation.

Primary care combined clinician and organisational questionnaire
The primary care combined clinician and organisational questionnaire will be sent for all children and young adults sampled for inclusion in the study where the GP can be identified. The questionnaire will be sent directly to the GP for completion either via the online questionnaire system, or as a hard copy questionnaire if the online system cannot be used.

5. Case note review
The case note review will be undertaken on the sample of children and young adults selected for inclusion in the study and will focus on a sample of people diagnosed with arthritis between 1st April 2019 – 31st March 2023. Notes will be requested from diagnosis until the 31st March 2023. If a child or young adult is identified and tracked across a number of healthcare settings, case notes will be requested from all organisations.

Notes requested will include:
From primary care

- All primary care notes which could relate to JIA including GP consultations, discharge summaries and follow-up letters

From acute, community and independent care

- GP referral letters
- Outpatient correspondence and clinic letters
- Consultation notes (including where available from electronic patient record systems)
- Discharge summaries for inpatient stays and outpatient appointments
- Therapy notes
- MDT summaries

- Outpatient prescriptions
- Imaging reports

Upon receipt at NCEPOD the case notes will be redacted if not already done so prior to sending.

Reviewer assessment form

A multidisciplinary group of reviewers (detailed below) will be recruited to assess the case notes and questionnaires and provide their opinion on what went well and what did not go well during the process of care via the reviewer assessment form.

Table 2 summarises the data sources for significant points along the pathway.

Area of enquiry	**Method of data collection**	**Confidentiality**
Primary care	Clinician questionnaire	Identifiable
	Online surveys and focus groups/interviews	Anonymous
Acute, community and independent care	Case notes, clinician questionnaire, organisational questionnaires	Identifiable
	Online surveys and focus groups/interviews	Anonymous

Sample Size

Data source	**Target number**
Young person online survey (non-identifiable)	50
Parent/carer online survey (non-identifiable)	50
Clinician online survey (non-identifiable)	300
Organisational questionnaire	~250
Hospital clinician questionnaires	Up to 15 per hospital
Case note review	Up to 15 per hospital

Analysis and Review of Data

Reviewers

A multidisciplinary group of reviewers will be recruited to assess the case notes and questionnaires and provide their opinion on what went well and what did not go well during the admission. The reviewer group will comprise rheumatologists (paediatric and adult), paediatricians, physicians, nurses, general practitioners, occupational therapists, physiotherapists, ophthalmologists, pharmacists, radiologists and orthopaedic surgeons.

An advert will be sent to Local Reporters to disseminate throughout the relevant departments. It will also be placed on the NCEPOD website and social media channels. Successful applicants will be asked to attend a training day where they will each assess the same two cases to ensure consistent assessment. A number of meeting dates will be arranged, and each reviewer will then be asked to attend a minimum of a further 4 meetings. NCEPOD staff will ensure there is a mix of specialties at each meeting from across the UK. Each meeting will be chaired by an NCEPOD clinical coordinator who will lead discussion around the cases under review. The meetings will either be held in person in the NCEPOD office, or over Microsoft Teams with secure and temporary access to the case notes for review (not downloadable or printable by the case reviewer). Towards the end of the study the reviewers will be invited to attend a meeting where the data will be presented to and discussed with them. The reviewers will also be sent two copies of the draft report for their comment as this is developed.

Confidentiality and data protection
All electronic data are held in password protected files and all paper documents in locked filing cabinets. As soon as possible after receipt of data NCEPOD will encrypt electronic identifiers and anonymise paper documents. Section 251 approval has been obtained to perform this study without the use of patient consent in England and Wales.

Ethical approval will not be required to undertake this study. Duty of candour is covered by the NCEPOD Cause for Concern policy, which ensure that any cases reviewed as less that satisfactory and as a cause for concern are discussed and action taken where required.

Study outputs
On completion of the study a report will be published and widely disseminated to all stakeholders to encourage local quality improvement (QI) (further details available in the communication plan). In addition to the report, supporting tools will be made available including:

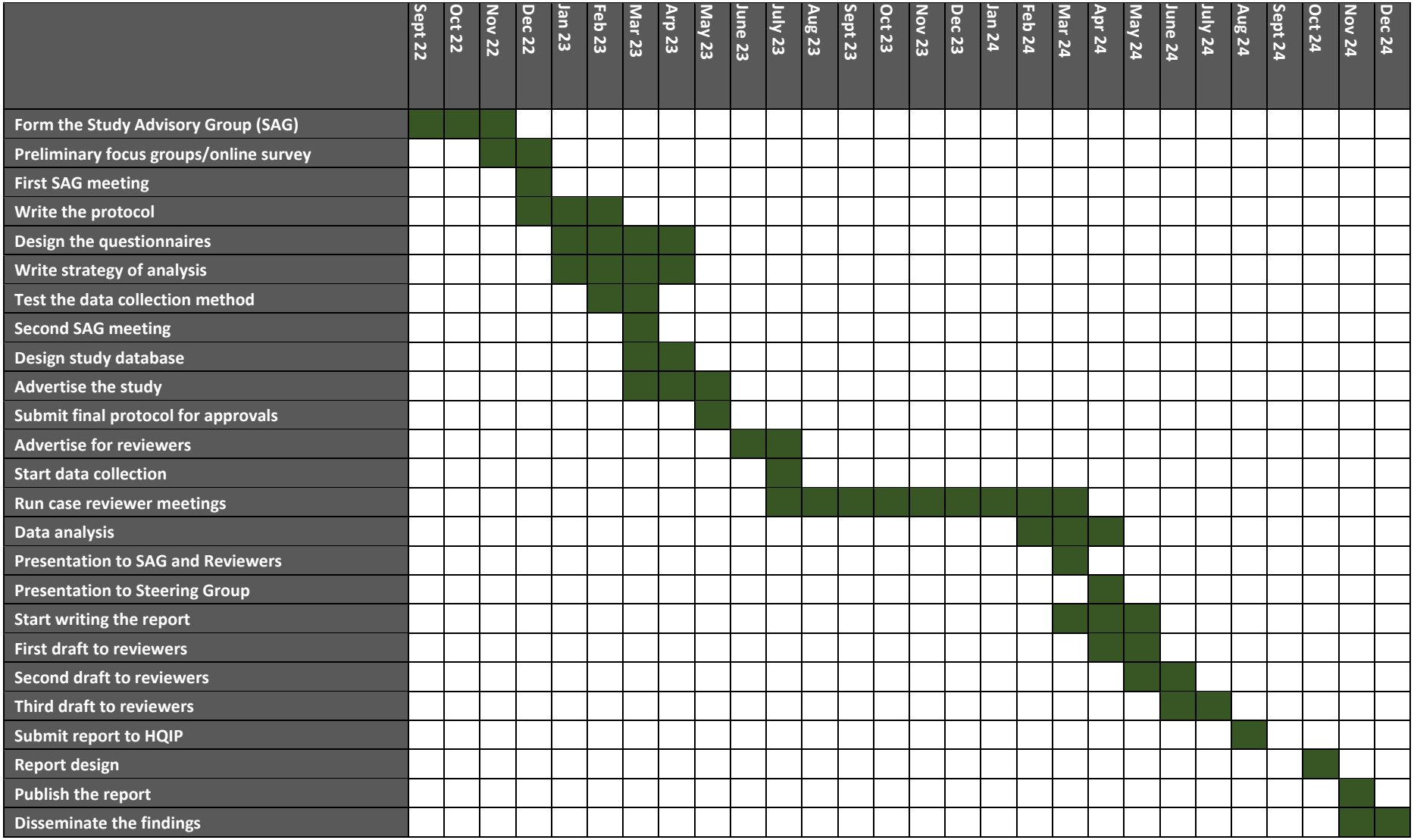
- A summary report and summary sheet
- Infographics
- The recommendation checklist
- An audit tool
- A slide set
- A guide for commissioners
- Quality improvement tools
- Useful links for children and young adults and parent carers

Examples of good practice will be shared, and additional QI tools will be developed where appropriate. Key messages from the report will be shared via social media.

Following publication, the report findings will be shared at national and local conferences, study days and other events; and papers submitted to journal for consideration for publication.

Data sharing
Post publication of the study there is the potential to share anonymised data sets with interested parties working in the same field. This will be undertaken following a strict process and will ensure the data does not become identifiable in their nature due to small numbers.

Timescale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Dec 24
Form the Study Advisory Group (SAG)
Preliminary focus groups/online survey
First SAG meeting
Write the protocol
Design the questionnaires
Write strategy of analysis
Test the data collection method
Second SAG meeting
Design study database
Advertise the study
Submit final protocol for approvals
Advertise for reviewers
Start data collection
Run case reviewer meetings
Data analysis
Presentation to SAG and Reviewers
Presentation to Steering Group
Start writing the report
First draft to reviewers
Second draft to reviewers
Third draft to reviewers
Submit report to HQIP
Report design
Publish the report
Disseminate the findings

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Appendix 1

Scoping focus group summary

At the study outset, three focus groups were undertaken to gather the views of young people and parent carers on what went well, and what did not go well with their Juvenile Idiopathic Arthritis to inform the direction of the study.

Participants were recruited via social media. Six parent carers and three young people participated in these sessions, with representation from across the UK.

All the parent carers' young people either themselves had JIA or cared for a young person with JIA. Their conditions ranged from mild to requiring a high level of care.

The areas the focus group participants indicated it was important to include in the study are listed below. These will feed into the development of the study aims and objectives.

Areas to review

- Delay to treatment and referral
- Age and cultural discrimination
- Lack of information on the side effects of methotrexate and other biological drugs
- Communication/relationship between specialties
- Lack of awareness from healthcare professionals about JIA
- Misdiagnosis
- Ongoing care plan
- Holistic care (support to get back into schooling and physical activities etc)
- The role of primary care