

## Concise report

# A survey to understand the feelings towards and impact of COVID-19 on the households of juvenile dermatomyositis patients from a parent or carer perspective

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### Abstract

**Objectives.** This aim of this study was to gain a better understanding of how parents and carers feel about the effects and impact of the coronavirus disease 2019 (COVID-19) pandemic lockdown and how this impacted upon their child/young person with JDM.

**Method.** We approached 139 participants from the JDM Cohort Biomarker Study (JDCBS), with specific consent to approach electronically for research studies. A secure electronic questionnaire with study introduction was sent to participants for their parents and carers around the UK to complete. It consisted of 20 questions about the impact of the pandemic on their child or young person's clinical care. Data were analysed quantitatively and qualitatively.

**Results.** There were 76 (55%) responses to the survey. More than 50% of participants were actively being treated for their JDM at the point of survey completion as recorded by their parent or carer. More than 40% attested to disrupted treatment owing to COVID-19. The biggest impact upon clinical care was cancellation of appointments, initiating virtual appointments and extension of time between blood tests. Parents and carers expressed their own feelings of worry, concern and anxiety, but also those of their child or young person.

**Conclusion.** Families who have a child or young person with JDM have been affected by COVID-19. Qualitative comments highlight that it has been a very difficult time. Further investigation is required into this area and could be compared with research on the effects of COVID-19 on other patient groups with chronic disease.

**Key words:** juvenile dermatomyositis, JDM, COVID-19, rheumatology, impact, feelings, sources, information, medication

### Key messages

- COVID-19 has disrupted the treatment of JDM.
- Parents/carers are worried, concerned and anxious about the effects of COVID-19 on their child/young person.
- Parents/carers had access to enough useful information to support their child/young person with JDM.

### Introduction

The coronavirus disease 2019 (COVID-19) pandemic and lockdown in the world is potentially a worrying time for everyone. The opinions of families that are caring for a child or young person with chronic disease need to be heard and addressed appropriately. The British Society of Rheumatology (<https://www.rheumatology.org.uk/practice-quality/covid-19-guidance>) and Versus Arthritis (<https://www.versusarthritis.org/covid-19-updates/informa>

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tion-for-children-and-young-people/) websites provide updated advice and information about how to manage and cope with a rheumatic disease during the COVID-19 pandemic for clinicians (BSR) and parents or patients in the UK (Versus Arthritis) [1, 2]. Across the Rheumatology community, many studies are being conducted to investigate the impact of COVID-19 on patients with rheumatic disease [3]. This study was designed to address the impact of COVID-19 specifically for the parents and carers of children or young people with the rare, autoimmune rheumatic condition, JDM. This study enabled assessment of how to support these families further in the future, especially if the pandemic continues.

## Methods

This study was designed for parents and carers of patients participating in the JDM Cohort and Biobank Study (JDCBS). This is a longitudinal, inception cohort study of children diagnosed with JDM, collecting clinical and biological data, with linked samples from diagnosis and serially over time [4, 5]. These specimens and clinical data sets enable collaborative research into JDM and other forms of juvenile myositis.

One hundred and thirty-nine participants from the JDCBS database were approached, with specific consent to contact electronically for research studies. An initial email was sent with information about the study, followed by an email with the link to the questionnaire with study summary (Supplementary Table S1, available at *Rheumatology Advances in Practice* online). The questionnaire link was sent out at the beginning of June 2020. The questionnaire was live for 4 weeks during 2020, during the first lockdown in the UK. The questionnaire was designed and managed through a secure University Software System compliant with data protection. On completion of the questionnaire, data were submitted electronically.

The demographics of the whole cohort of JDCBS participants were compared between those sent the questionnaire and those not sent the questionnaire, to exclude bias in the survey population. Data were described using counts, percentages, medians and interquartile range (IQR), with Student's unpaired *t*-test used to calculate significance. Demographics of the responders and non-responders to the survey were compared to exclude bias. Data were described using counts, percentages, medians and IQR, with the Mann-Whitney *U*-test used to calculate significance. Clinical data used were kept minimal and were self-reported by the parents and carers. Free text responses were depicted as quotes, word clouds and graphical bar charts. Analysis was a ranking system of the number of times a word was recorded in the total answer set, assigned a font size according to scale and represented as a word cloud.

Parents and patients gave written informed consent or age-appropriate assent. The study obtained ethical approval through North-East Yorkshire Research Ethics Committee (MREC, 01/3/022) for JDM sample collection.

All consent was obtained in accordance with the Declaration of Helsinki.

## Results

### Demographics of cohort

Responses were received from 76 (55%) participants who were sent the survey. Eleven (15%) of the surveys were recorded as completed by the young person with JDM. For the majority of the analysis, the responses have been shown as a combined data set of all responses irrespective of the responder. Participants with email addresses who were sent the survey had comparable overall demographics to those not sent the survey (given that no email address had been stored with consent to re-contact; Table 1). The median current age at survey was significantly lower for those participants sent the survey, at 18 years, compared with 20.53 years for those who were not sent the survey (no email;  $P=0.0013$ ). The median duration since diagnosis was significantly lower for participants who were sent the survey, at 9.2 years, compared with the remaining participants in the JDCBS, at 12.1 years (without email;  $P=0.0001$ ). These differences can be explained, because the JDCBS started to collect emails only recently, and not from the start of the study. The ethnicity demographics were also comparable, with no obvious differences found. There were no significant differences in these demographics between responders and non-responders to the survey (Supplementary Table S2, available at *Rheumatology Advances in Practice* online).

### Impact of COVID-19 on JDM treatment

More than 50% of participants were actively being treated for their JDM at the point of survey as recorded by their parent or carer. Actively treated was defined by, at point of survey, the patient being reported to be taking medication at the time of survey completion. Of the responses, 45 (59.2%) recorded that the child or young person with JDM was taking medication at the point of survey and that 42 (55.3%) had an appointment in 2020. Of those on medication, 10 (24%) were taking prednisolone, 29 (69%) MTX and 26 (62%) HCQ (full data are in Supplementary Table S3, available at *Rheumatology Advances in Practice* online). Participants were asked to record the points of disruption to their child or young person with JDM treatment. Thirty-one (40.8%) responses attested to disrupted treatment during 2020, of which the main causes were appointment cancellations, initiating virtual appointments and an extension of time between blood tests (Fig. 1A). Only six patients were admitted to hospital for 'day case for infusion', one 'overnight or extended' and two 'other' since lockdown began in March 2020.

### Overall feelings about COVID-19

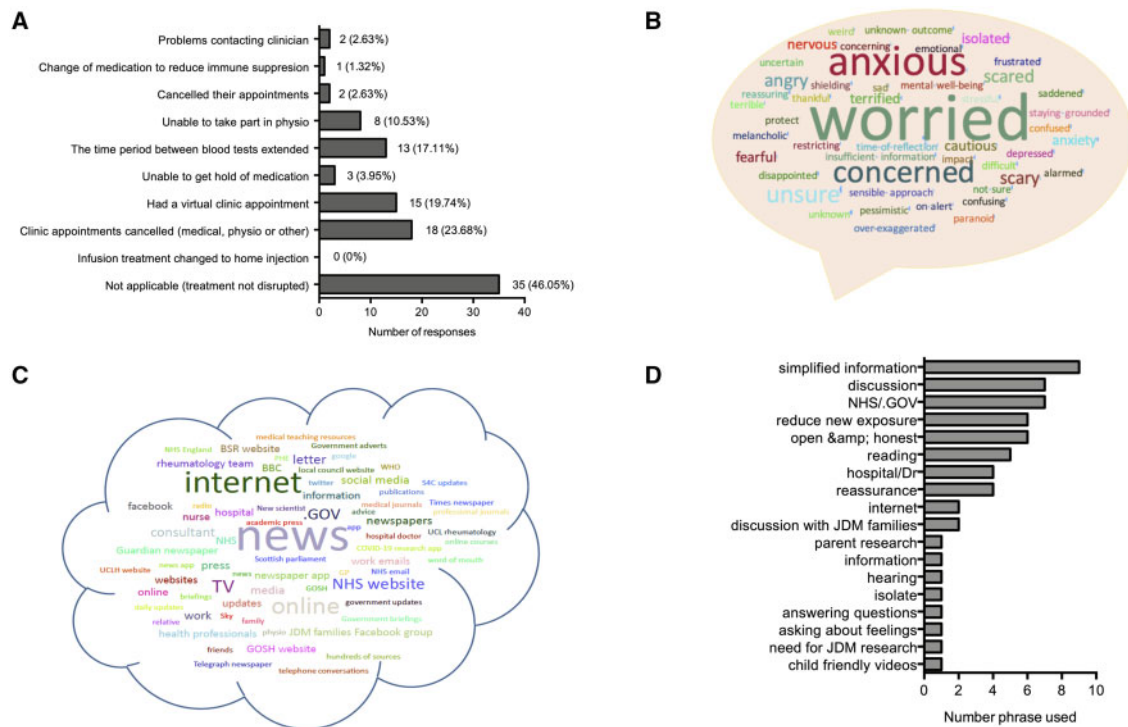
A set of questions in the survey described the feelings of the parents and carers about COVID-19. Question 10 was answered as a maximum of five-word descriptive free text, enabling the participant to answer freely. Quotes include: 'Scary cautious sensible approach'; 'Worried, anxious,

**TABLE 1** JDM Cohort Biomarker Study participant demographics comparison of those who were sent the survey and those who were not

| Characteristic   | JDM cohort                      |                                     |
|--|---------------------------------|-------------------------------------|
|  | Questionnaire sent<br>(n = 136) | Questionnaire not sent<br>(n = 454) |
| Age at diagnosis, median (IQR), years; <i>P</i> = 0.9974     | 7.57 (4.60–10.75)               | 7.48 (4.81–11.00)                   |
| Current age, median (IQR), years; <i>P</i> = 0.0013          | 18 (12.37–22.97)                | 20.53 (14.59–26.39)                 |
| Time since diagnosis, median (IQR), years; <i>P</i> = 0.0001 | 9.14 (5.14–13.72)               | 12.05 (6.97–17.55)                  |
| Female sex, <i>n</i> (%)                                     | 90 (66.18)                      | 323 (71.15)                         |

Abbreviations: IQR: interquartile range.

**FIG. 1** Responses to descriptive questions



(A) The main causes of disruption to JDM treatment responses by multiple choice; data are described as the number of responses and percentage of the overall number of responses. (B, C) Free text responses on overall feelings about COVID-19 (B) and sources of COVID-19 information (C), with analysis by a ranking system of the number of times a word was recorded in the total answer set, assigned a font size according to scale and represented as a word cloud. (D) Sources of information on COVID-19 accessed and how this information was translated to their child or young person with JDM, with analysis by a ranking system of the number of times a word was recorded in the total answer set and represented as a bar chart.

depressed, fearful'; and 'Anxiety heightened by son's JDM'. The main feelings about COVID-19 recorded were worried, concerned and anxious (Fig. 1B).

**Where, what and how information about COVID-19 was obtained**

To comprehend the accessibility and usefulness of information regarding COVID-19, we asked the participants what their sources of information were and how they

relayed the information to their child or young person with JDM. Most responders recorded that they had access to multiple sources of information about COVID-19, including the news, Internet and the National Health Service/BSR websites (Fig. 1C). The most common modality of relaying information about COVID-19 to their child or young person was family discussion, simplifying information and being honest about the situation (Fig. 1D). The 11 young people with JDM who completed the survey responded that their main sources of

information on COVID-19 were from their own research. Quotes from parents and carers include: 'We have discussed it but not in depth and not with particular emphasis on JDM'; 'I try to make sure they are hearing and reading factual information that is suitable for their ages'; 'It's hard to find something age appropriate'; and 'We share the information we feel is age appropriate'. Fifty-four (71.1%) participants said the information sourced was useful, and 43 (56.6%) said they had access to enough information to support their child or young person. When asked what impact COVID-19 had on their child or young person with JDM, 32 (41.6%) said that COVID-19 had had a negative impact; this was a multiple choice question, and the other options were positive or indifferent impact.

### COVID-19 symptoms

Finally, we asked for information about COVID-19 directly. Only 13 (17.1%) reported a family member having had symptoms of COVID-19, of whom 6 (7.9%) were the child or young person with JDM, and only 2 (2.6%) recorded a positive test result of a family member.

## Discussion

This survey was conducted in a small cohort with a rare disease to address the feelings towards and impact of the COVID-19 pandemic. The survey was completed by the parents or carers of a child or young person with JDM who is a participant in the JDCBS. To our knowledge, this is the first study of its kind in this patient group and is therefore a starting point to consider how concerns about COVID-19 impact upon children and young people with a rare autoimmune condition. Recently, Livermore *et al.* [6] highlighted how children and young people with JDM have expressed feelings of confusion, uncertainty and a feeling of difference from their peers. Adding to this, anxiety over a worldwide pandemic, in which children and young people are isolated from their schools and usual support systems, could cause serious concerns.

This survey shows that families with children and young people with JDM have been adversely affected by COVID-19. They have experienced disruption to treatment and feel that COVID-19 has had a negative impact on the child or young person with JDM. They are worried, concerned and anxious about the effects of COVID-19, but feel that they have had access to enough useful information to support their child or young person with JDM. These data support the findings of the Juvenile Arthritis Research (JAR) study, which showed that participants in the registry had high levels of worry, but parents were more worried than adult patients [3]. Given that this study is longitudinal, they have also reported that currently the level of worry is reducing each week, although this might change as the pandemic continues.

## Limitations

One of the limitations of this study was that the questionnaire was designed for the parent or carer to complete, and therefore the results were from their perspective rather than the patient; however, 11 out of the 76 responders were, interestingly, the patients themselves. We are aware of an extensive literature that suggests discrepancies between parents and young people are common in this type of data, and a limitation of this study is that we did not have paired responses from both parents and their child/young people to explore this issue [7, 8]. In this study, we noted the transfer of face-to-face appointments to virtual appointments but did not assess the opinion of the parent/carer or child/young person of this change; this would be useful to explore in the future. Our study used JDM as the terminology for this disease cohort, but we are aware that a small percentage of the cohort have other forms of idiopathic inflammatory myopathies. We believe that as an assessment of chronic disease, the results were not affected by this terminology. This survey did not address the impact of the COVID-19 pandemic on exercise and education, but these would be important topics for further investigation. This was a cross-sectional study, with answers from the participants at the point of survey only, not longitudinal, and therefore reflect the beliefs at the time of survey. Forty-five per cent of participants did not respond to the survey, which is less than average for survey non-response [9]. We can only speculate at the reasons for this. At this anxious time, research might not be a priority for these families. Home schooling, working from home, shielding and altered care patterns could be some examples of why these participants did not respond.

## Conclusion

This survey has provided initial information on the effects of COVID-19 on patients with JDM and their families. The data support the findings from a Hong Kong study, which reported the impact of COVID-19 pandemic on patients with rare diseases [10]. Although not specific to paediatric patients, they described that the COVID-19 pandemic had impacted the health status (46%), service use patterns (71%) and mental health (76%) of the patients. There are many survey studies across the world that are collecting data on the impact of the COVID-19 pandemic on rheumatology, rare disease and paediatric patients (<https://rheum-covid.org/patient-survey/>) [3]. Other studies include the ImmunoCOVID19 study that is monitoring COVID-19 infections in children and adults possibly more vulnerable to infections (<https://www.uhs.nhs.uk/ClinicalResearchinSouthampton/Public-and-patients/Featured-research-studies/Featured-research-ImmunoCOVID19-study.aspx>) and the Versus Arthritis Centre for Adolescent Rheumatology survey assessing the impact of COVID-19 on JDM and JSLE from an adolescent patient perspective (<https://redcap.slms.ucl.ac.uk/surveys/?s=8RHL3TXCXD>) [11]. These data sets will improve the understanding, support

and management of rheumatic disease during this pandemic. The COVID-19 pandemic is still on-going and affecting the treatment and management of chronic disease. The uncertainties during this time need to continue to be addressed in order that we can adapt the care and support for these patients. These elements need to evolve to cope with the long-term effects of the pandemic.

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## Data availability statement

The authors confirm that the data supporting the findings of this study are available within the article (and/or) its [supplementary materials](#). Raw data were generated under the auspices of the JDCBS, stored securely at UCL Great Ormond Street Hospital Institute of Child Health. Derived data supporting the findings

of this study are available from the corresponding author (M.G.L.I.W.) on request.

## Supplementary data

Supplementary data are available at *Rheumatology Advances in Practice* online.

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