

# The COVID-19 Pandemic Highlights An Unmet Need To Improve Remote Assessment of Disease Activity in Rare Diseases, Juvenile-Onset Systemic Lupus Erythematosus And Juvenile Dermatomyositis

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## Short Report

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# Abstract

Here we report the results of a pilot young patient survey which targeted patients with juvenile systemic lupus erythematosus (JSLE) and juvenile dermatomyositis (JDM), which explored wellbeing, resilience and general concern about the COVID-19 pandemic. The survey was completed anonymously by patients who have been approached via the automatically generated hospital data base between June-December 2020. This pilot study found that generalised concerns regarding the COVID-19 pandemic were significantly higher in females, although their self-reported disease activity scores were comparable with male respondents. Our findings highlight a need to improve remote assessments to facilitate triaging for face-to-face visits in the situation of future pandemics associated with disruption of clinical services, as well as further research in this area of young patient-reported outcomes outside hospital visits.

## Background:

The impact of the COVID-19 pandemic on the delivery of healthcare for adolescents and young people should not be underestimated, even though this population is less clinically vulnerable to COVID-19 infection. Physiological, psychological and social-role changes during this period heavily influence future well-being and health. The additional consequences of disrupted access to education, work and healthcare, and social and financial stresses during the pandemic have had significant impact on mental health and well-being(1). Young people with chronic health conditions, already face a disproportionate psychosocial burden compared with their peers, and are likely to face additional worries and concerns regarding COVID-19, including heightened health-related anxiety, disrupted routines, reduced access to physical and psychosocial support(2).

As clinicians working in a specialist centre, we aimed to evaluate the impact of the COVID-19 pandemic in adolescents and young adults with juvenile-onset systemic lupus erythematosus (JSLE) and juvenile dermatomyositis (JDM). These are rare, complex autoimmune connective tissue disorders with a female preponderance, associated with multiple organ involvement and significant impact on quality of life. Here we report the results of a pilot survey, which explored wellbeing, resilience and concern about the COVID-19 pandemic in patients with JSLE and JDM.

## Patient Cohort And Methods:

The survey was completed anonymously by patients between June-December 2020. The hospital digital database generated a complete list of patients with JDM and JSLE, who were under the care of Department for Adolescent Rheumatology at University College London Hospital in 2020. Automatic emails were sent to registered patient emails available in the database and patients of age 16 and over (who were legally able to consent at the time of inclusion in this exploratory survey) were invited to complete the survey anonymously accessed via a link to a RedCap questionnaire database. None of the patients have been approached to complete the questionnaire when attending clinics in person to

minimise the potential selection bias. The survey has been approved by the local ethics committee (Ref. 20/HRA/2565).

Results were analysed using descriptive statistics (using R). The survey (available as Supplementary file) contained questions about patient's demographics (age, ethnicity, gender), employment and education status, disease duration, age at onset, as well as well-being or resilience, using the validated mood and feeling questionnaire (MFQ)(3), resilience scale(4) and the Warwick-Edinburgh mental well-being scales(5). The patients were also required to complete a VAS 0-100 scale to rate their general concerns regarding the general impact of COVID-19 on their lives.

## Findings:

There were 63 respondents (53 females; 10 males) who answered the survey during the recruitment period. The survey had a 47% response rate, which is not unsurprising considering that patients have not been approached in person as many of the routine clinical assessments were conducted virtually. Reflecting the significant gender bias in JSLE and the higher prevalence of JSLE compared to JDM, there was a predominance of young females completing the survey (84%). There were no significant gender differences in the disease duration, age at diagnosis, disease duration, age of responders or immunosuppressive medication according to self-reported gender (Table 1). The majority of patients were living outside London as we are a tertiary centre for adolescent and young adult rheumatology. Generalised concerns regarding the COVID-19 pandemic were significantly higher in females ( $p=0.007$ ), although their self-reported disease activity scores were comparable with male respondents ( $p=0.205$ ) [Table 1, Figure 1A]. The self-reported resilience was almost identical in young men and women ( $p=0.99$ ).

Linear regression analysis, adjusted for age, ethnicity and gender demonstrated that self-reported disease activity (using a global visual analogue scale) was the strongest determinant of concerns associated with COVID-19 ( $p<0.0003$ ) (Figure 1B), which was evident in both females ( $p=0.003$ ) and males ( $p=0.004$ ). Longer disease duration was associated with general concerns regarding COVID-19 pandemic in males only ( $p=0.018$ ). There was no association between COVID-19-associated concerns and employment and education status, or well-being or resilience, using the validated mood and feeling questionnaire (MFQ)(3), resilience scale(4) and the Warwick-Edinburgh mental well-being scales(5).

## Conclusion:

To our knowledge, this is the first focused evaluation of the impact of the COVID-19 pandemic on patients with JSLE and JDM. Our results demonstrate that gender differences should be considered when assessing patients, and that concerns regarding COVID-19 strongly correlate with patient-reported disease activity when accounting for other confounders. We were unable to compare patient-reported and physician-assessed disease activity, given restricted access to clinic appointments during the height of the pandemic. Previous psychology research has identified gender differences in wellbeing in healthy

young people(6), while the most important predictors of well-being in emerging adulthood were higher self-confidence and lower negative self-evaluation(7), aspects that are likely to be affected by having to live with a chronic condition such as JSLE or JDM. In addition, well-being at the age of emerging adulthood is recognised as having significant impact on further psychological well-being (8); therefore addressing additional psychological stressors, such as general concern related to COVID-19 pandemic in young people with chronic conditions could have long term benefits.

The main limitations of this exploratory survey are related to the relatively small sample size of this single centre study, which does not allow for generalization of results, and inability to establish any causal relationship between the outcomes measured. There are still uncertainties about the outcomes of COVID-19 infection/vaccination in children and young people with chronic inflammatory conditions, despite the re-assuring signals to date (9-12); this is likely to be reflected in their general COVID-19-associated concern.

Telemedicine gained traction during the pandemic and is likely to have a more permanent place in mainstream healthcare provision hereafter. Virtual consultations are particularly relevant for continuity of routine care for rare diseases in high-risk groups, including our cohort of JSLE and JDM, as patients often have to travel considerable distances to obtain tertiary expertise and may be shielding due to inherent and iatrogenic immunocompromise. There are recognised advantages and disadvantages of care delivery via virtual consultations for young people (13, 14), in addition to the challenges of suffering from a complex autoimmune condition such as JSLE and JDM which have to be taken into account when designing virtual clinical services. Our findings highlight a need to improve remote assessments to facilitate triaging for face-to-face visits, by developing standardised composite patient reported outcome measures for disease activity. Emerging digital technologies for self-assessment, monitoring and reporting of disease activity are likely to be important tools to empower young people and promote independence, as they transition to young adult services.

## Abbreviations

JDM- juvenile dermatomyositis

JSLE – juvenile systemic lupus erythematosus

MFQ - mood and feeling questionnaire

VAS – visual analogue scale

## Declarations

**Ethical Approval and Consent to participate:** The survey has been approved by the local ethics committee (Ref. 20/HRA/2565).

**Consent for publication:** was included in the ethical approval.

**Availability of supporting data:** supplementary data can be made available upon request

**Competing interests:** all authors declared no conflicts of interest.

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**Authors' contributions:** CC designed the questionnaire with input from AK, W-HW, PM, LH, GD, PH, JH, EJ. JP performed the data analysis with input from EJ, CC. All authors contributed to data collection and reviewed the data analysis. JP, PM and CC wrote the paper. All authors reviewed and approved the final manuscript.

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**Authors' information:** not further relevant information.

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## Tables

**Table1:** Patient characteristics according to self-reported gender

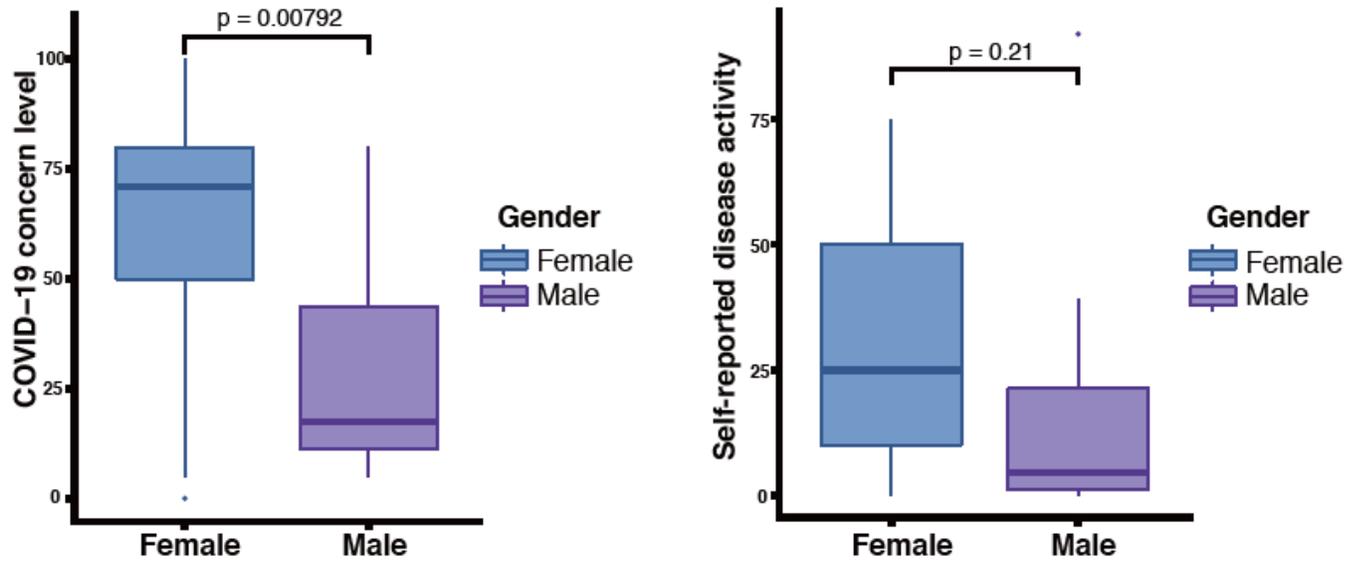
	Female	Male	p-value
<b>Number</b>	53	10	-
<b>Current age mean (IQR)</b>	22.42 (19.00-27.00)	22.86 (17.15-29.25)	0.87
<b>Age at diagnosis years mean (IQR)</b>	11.47 (9.00-14.00)	10.00 (8.00-11.75)	0.26
<b>Disease duration; Mean (IQR)</b>	10.94 (5.00-16.00)	12.86 (9.70-18.75)	0.46
<b>Ethnicity;</b>			
White	33 (62.3%)	8 (80.0%)	0.47
Non-white	20 (37.7%)	2 (20.0%)	
<b>Location;</b>			
London (%)	13 (24.5%)	2 (20.0%)	1.00
Other (%)	40 (75.5%)	8 (80.0%)	
<b>Immunosuppressive treatment (%)</b>	40 (75.4)	8 (80%)	0.97
<b>Prednisolone treatment (%)</b>	22 (41.5)	5 (50%)	0.83
<b>Well-being (VAS 0-5) mean (IQR)</b>	3.066 (2.500 – 3.500)	3.486 (3.304 – 4.143)	0.27
<b>Resilience (VAS 0-7) mean (IQR)</b>	4.764 (3.714 -5.571)	4.757 (3.929 – 6.161)	0.99
<b>Self-reported disease activity (VAS 0-100) mean (IQR)</b>	30.83 (10.00-50.00)	17.70 (1.00-21.25)	0.205
<b>Self-reported COVID concern (VAS 0-100) mean (IQR)</b>	61.77 (50.00-80.00)	30.90 (11.25-43.75)	0.007
<b>Still in education</b>			
No	27 (50.9%)	5 (50.0%)	1.00
Yes	26 (49.1%)	5 (50.0%)	
<b>From those previously employed, currently working</b>			
No			

Yes	12 (44.4%)	0 (0%)	0.24
	15 (55.6%)	4 (100%)	

Legend: IQR- interquartile range; VAS – visual analogue scale.

## Figures

**A**



**B**

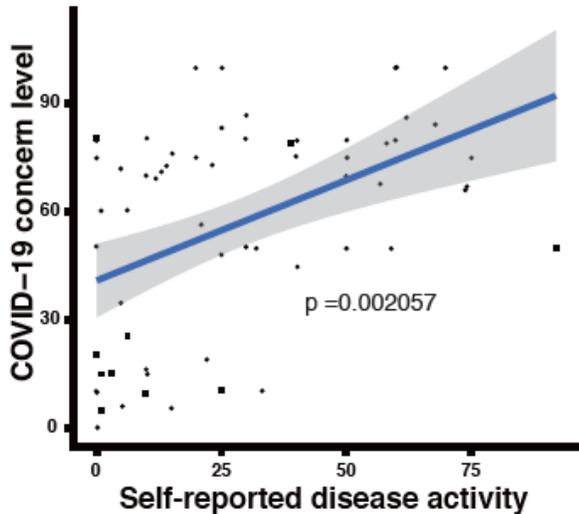


Figure 1

A First panel - Differences in COVID concern level (VAS 1-100, 100 being the highest concern regarding health-outcomes during COVID-19 pandemic) according to gender in patients with JSLE and JDM Second

panel - Differences in self-reported disease activity (VAS 1-100, 100 being the highest self-perceived disease activity) according to gender in patients with JSLE and JDM B Linear regression analysis, adjusted for age, ethnicity and gender demonstrated that self-reported disease activity (using a global visual analogue scale) was the strongest determinant of concerns associated with COVID-19 pandemic.

## Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- [Surveyblank.pdf](#)