

Modern Rheumatology

Social support and its relationship with health-related quality of life in patients with antiphospholipid (Hughes) syndrome --Manuscript Draft--

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| Manuscript Number: | MORH-D-16-00524R1 |
| Full Title: | Social support and its relationship with health-related quality of life in patients with antiphospholipid (Hughes) syndrome |
| Article Type: | Original Article |
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| Abstract: | <p>Objectives Antiphospholipid syndrome (APS) is recognised as a systemic autoimmune disease defined by recurrent thromboembolic events and/or pregnancy morbidity. Little is known about the psychological burden of this long-term condition. This study aims to explore the relationship between social support and health-related quality of life (HRQoL) in patients with APS.</p> <p>Methods 270 patients with a clinical diagnosis of APS participated in a cross-sectional online questionnaire survey collecting data on demographics, disease-related information, social support and HRQoL.</p> <p>Results Both perceived and ideal social support were associated with HRQoL in APS. Patients reported receiving insufficient social support. Perceived emotional support was related to physical functioning (B=7.77;95%CI:2.25,13.29); perceived instrumental support with bodily pain (B=17.52;95%CI:11.15,23.90) and perceived informational support with physical and social functioning (B=-6.30;95%CI:-12.52,-0.08; and B=8.06;95%CI:1.17,14.94). Ideal emotional support was related to physical and social functioning (B=5.80;95%CI:0.26,11.34; and B=7.53;95%CI:0.55,14.51); ideal instrumental support was associated with mental health (B=4.73;95%CI:0.38,9.07) and ideal informational support with vitality (B=5.85;95%CI:1.23,10.46).</p> <p>Conclusions Social supported was linked to HRQoL in patients with APS. Insufficient social support was associated with limitations in various HRQoL domains. Patient-tailored interventions addressing psychosocial aspects of living with APS might need to be incorporated in the treatment regimen to improve patients' psychological and physical status.</p> |
| Response to Reviewers: | Reviewer 1 Major comments: 1. The authors used short-form health survey (SF)-36 to assess HRQoL and a questionnaire to evaluate the social support. Multiple scales were used to score six of the eight domains in SF-36. How the other two domains were scored? There is no |

information in this manuscript indicating how the support survey values were computed in the social support questionnaire.

Thank you for your very useful suggestion. Indeed, it was unclear how the remaining two domains were scored on the SF-36. This has now been amended to read: "Multipoint scales (3 to 10 items) are used to score six of the eight domains while SF and BP are scored on a two-point scale (yes/no)" (page 7 lines 24-27). Similarly, scoring of the social support questions is not adequately described. Thank you for drawing our attention to this. Scoring scale details have now been added to the sentence referring to the social support questions description to make it clearer to the reader as follows: "The survey questions were presented in the form of a set of multiple choice questions with 4 or 5 possible options displaying various examples of social support scored on a two-point scale. The participants had to indicate which of the available support options listed (e.g. for emotional support: (a) listening; (b) understanding; (c) encouragement; (d) positive feedback; (e) willingness to learn more about the illness) they felt they were receiving (perceived support) and which one(s) they would like to still be receiving (ideal support) by simply ticking the appropriate answer yes or no (please see Appendix 1) (page 7 lines 1-8).

2.The classification in physical and mental domains in the SF-36, as analyzed in the tables, should be defined in the methods section.

Apologies for the omission. You are absolutely right. The physical and mental components of the SF-36 should have been specified in the methods section. This has now been added to the paragraph that describes the SF-36 domains as follows: "Specifically, the SF-36 measures eight domains relative to physical and psychological status which are termed "physical components" (PC) and "mental components" (MC). The physical components include: role physical (RP), general health (GH), bodily pain (BP), and physical functioning (PF) while the mental components encompass: role emotional (RE), vitality (VT), mental health (MH), and social functioning (SF) (page 7 lines 14-25).

3.According to the authors 270 out of 443 patients completed the survey. Four questionnaires were discharged. Is the final number of participants 270 or the number became 266 after discharging four questionnaires?

Apologies for the confusion. The total number of included questionnaires was 270 after discarding the 4 incomplete ones. This has now been clarified and reads: "274 patients completed and returned the questionnaire survey out of a total of 443 individuals who were sent the questionnaire survey link. Four questionnaires were discarded due to insufficient data which resulted in 270 completed questionnaires being included in the analyses" (page 8, lines 50-53).

4.Results of the SF-36 are presented as mean scores more or less than 60 (page 8). The authors should explain why the level to classify scores was set up at 60 and not at 50 or 80 for example.

This is an excellent point! Thank you for emphasizing the lack of explanation for the choice of the cut-off score. This has now been explained and reads as follows: "Mean scores in six domains (RP, BP, GH, VT, SF and RE) were <60 which is the cut-off score reported to indicate highest specificity for functional limitations [51]".

5.The computed values for the results obtained from the support questionnaire, perceived and ideal, have to be displayed in the results section.

Thank you for the suggestion and apologies for the omission. Computed values for the results obtained from the perceived and ideal social support questionnaire have now been added as text following participant characteristics in the results section pages 9/10 as stated below and results are also presented in an additional table (Table 1, page 25).

"Social support

Frequency statistics were computed separately for actual and idea social support measures. The majority of patients indicated that they were perceived to be receiving emotional support such as listening (78.7%), understanding (59%), encouragement (55.6%) but not positive feedback (34% vs 66%) and willingness on behalf of their

family and friends to learn more about APS (44% vs 56%). In contrast, the majority of patients perceived not to be receiving instrumental support such as help with childcare and housework/shopping (85.1% and 51.9% respectively), provision of transportation (69.8%), financial help (73.5%) and someone to accompany them to GP and hospital appointments (51.5%). Most of the information support, APS patients perceived to be receiving was derived from the internet (82.1%) and support groups (65.7%) whereas a minority of patients reported perceived informational support obtained from GPs (26.9%) and TV or leaflets (14.2%) (Table 1). The main social support (ideal) that patients would like to receive were: understanding (67.9%), willingness to learn more about APS from family or friends (64.9%), information from GPs (74.3%), TV and leaflets (50%) (Table 1)."

6. In this study, the authors did not present any subgroup analysis based in the clinical diagnosis of the patients (primary APS and APS associate with other conditions-SAPS). Therefore, the second paragraph of the discussion referring to the differences in the scores between primary APS participants and those with APS associate with other diseases should be omitted.

Excellent point and thank you for drawing this to our attention. The paragraph describing differences in scores between PAPS and SAPS patients has now been removed from the discussion section.

7. The discussion section should be enriched by adding potential strategies to incorporate the social support into the managements of patients

Very good point! It would indeed be helpful for the reader to provide specific potential strategies to incorporate social support into the management of patients. This has now been added and reads: "Specific strategies could be implemented through both primary and secondary care and include patient- and family/friends-education sessions delivered by specialist nurses. These sessions could provide disease- and treatment-specific information and self-management strategies such as International Normalised Ratio (INR) measuring, dietary advice and pacing to patients as well as disease-related information and ways of supporting their loved one in coping more effectively with APS to families/friends of patients with APS."

Minor comments:

1. Page 6, last paragraph should be moved from methods and presented in the discussion section.

Thank you for the suggestion. Last paragraph of page 6 on the HRQoL measure has been moved from methods section to discussion as suggested.

2. The sentence indicating that the researchers sent a reminder to complete the survey is duplicated in page 6.

Duplicated sentence indicating that the researchers sent a reminder to complete the survey was removed from page 6 (lines 36-38).

3. In page 11, the meaning of the last sentence is unclear.

You are absolutely right and thank you for drawing our attention to this point. Last sentence on page 11 "higher need to be listened to was found to be associated with better physical functioning" was rephrased to read "People who felt they still needed someone to listen to their concerns and worries would be more likely to have better physical functioning if they had this support".

4. Tables. The statistical method used for the analysis have to be indicated in the footnote.

Apologies for the omission. Tables 1-6 now have an added footnote indicating the statistical analysis performed.

5. All abbreviations should be defined at first mention and used consistently thereafter (SF-36, GPs)

Excellent point! Thank you for emphasizing this. All abbreviations have now been defined at first mention and used consistently thereafter [(SF-36), General Practitioner (GP), Hughes Syndrome Foundation (HSF)] etc.

Reviewer 2

Major points:

1. In your manuscript, you assess the association between ideal social support and HRQoL. However, as you mentioned in Introduction, ideal social support is less important than perceived support for QoL of the patients with APS. You should describe the reason why you assessed the relationship of ideal support with HRQoL in Introduction or Discussion.

This is an excellent point and we thank you for drawing it to our attention. We have now added a paragraph in the introduction (page 5, following the sentence "In addition, lack of understanding from their environment regarding their illness and its consequences contributes to poorer adjustment to chronic illness" explaining the reasons why we included ideal social support in our study stating "In the present study we explored both forms of social support because we attempted to evaluate the magnitude of discrepancy of perceived and ideal social support in patients with APS and whether this discrepancy was associated with their HRQoL. Furthermore, we wanted to assess how much social support patients with APS feel they still need compared to how much they feel they are receiving in order to be able to provide them with additional support to improve their HRQoL".

2. Furthermore, you should discuss the interpretation and clinical means of your results of the analysis for the association between ideal social support and HRQoL (Table 4, 5, 6).

Thank you for your extremely helpful suggestion. Discussing the interpretation and clinical means of our results of the analysis for the association between ideal social support and HRQoL is indeed vital in order to highlight the importance of our findings and implications. A paragraph has now been added in the discussion section (page ...) discussing the association between ideal social support and HRQoL, its meaning and clinical implications for patients with APS reading: "Similarly, higher ideal social support was also linked to better HRQoL. People who felt they needed someone to listen to their concerns and worries were predicted to be more likely to have better physical functioning such as fewer limitations in performing physical activities, including bathing or dressing and experience greater vitality i.e. feeling more energetic. This could be attributed to the fact that people might be likely to receive some reassurance from their social circle that they are able to perform certain activities following the expression of their concerns or offer them additional support which might contribute to decrease the burden that affect their energy levels. This was also reflected in the fact that the need for more frequent encouragement was associated with better physical while greater provision of positive feedback was associated with better role physical greater vitality, and role emotional.

The need to feel understood by family and friends was linked to better HRQoL in all domains except for physical pain and role emotional. Indeed, negative social responses, particularly discounting (rejecting) and lack of understanding (not being acknowledged), were associated with poorer health among patients with fibromyalgia and rheumatoid arthritis (Kool et al., 2010). Greater understanding would be derived from better knowledge of APS by the patient's family and social circle. Results showed that patients' greater need for their family and friends to learn more about APS was related to better role physical, and greater vitality. Ideal instrumental support was associated with better HRQoL in all domains in terms of provision of help with housework and/or shopping and attendance at GP and hospital appointments and greater informational support was associated with better role emotional and better mental health, while information from support groups was associated with greater vitality.

Minor points:

1.Results: Relationship between social support and HRQoL in APS, line 1-3, this paragraph should be described in Material and Methods.

The sentence "Multiple linear regression analysis was performed to explore associations between social support and HRQoL in patients in APS and to examine whether perceived and ideal social support were associated with HRQoL" (line 1-3 in results) has now been moved to the last paragraph of the Material and Methods section.

2.Results: Perceived social support and HRQoL, line 4, you described that your result showed higher levels of encouragement were related to "poorer" role physical functioning. I think "better" is correct.

You are absolutely right and my sincere apologies for the typographical error. "Poorer" role physical functioning has now been corrected to read "better" role physical functioning.

3.Results: Discrepancy values between perceived and ideal social support, line 10, you described it would seem that the "emotional" supports APS patients perceived to be far smaller than the support they ideally would need to be receiving. I think "Informational" support is correct.

This is an excellent point! Thank you for drawing our attention to this error. The sentence "it would seem that the "emotional" supports APS patients perceived to be far smaller than the support they ideally would need to be receiving" has now been amended to read "the "informational" support APS patients perceive to be receiving is far smaller than the support they ideally would need to be receiving".

4.Discussion: line 14, you described that higher "provision" of instrumental support "provision" was related to better HRQoL (...). Second "provision" is supposed to be extra word.

Thank you for drawing our attention to this duplication error. Second reference to "provision" has now been removed from the sentence.

1 **MORH-D-16-00524 Received: 9-Sep-2016; Accepted: 30-Mar-2017**

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4 **Original Article**

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10 **The relationship between social support and health-related quality of life in patients with**
11 **antiphospholipid (Hughes) syndrome**

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1 **Abstract**

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3 **Objective.** Antiphospholipid (Hughes) syndrome (APS) is recognised as a systemic autoimmune disease
4 defined by recurrent thromboembolic events and/or pregnancy morbidity. Little is known about the
5 psychological burden of this long-term condition. This study aims to explore the relationship between
6 social support and health-related quality of life (HRQoL) in patients with APS.
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12 **Methods.** 270 patients with a clinical diagnosis of APS participated in a cross-sectional online
13 questionnaire survey. Data included: demographics, disease-related information, social support and
14 HRQoL.
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19 **Results.** Both perceived and ideal social support were associated with HRQoL in APS. Patients reported
20 receiving insufficient social support. Perceived emotional support was related to physical functioning
21 (B=7.77, $p=0.006$, 95% CI: 2.25, 13.29); perceived instrumental support was associated with bodily pain
22 (B=17.52, $p<0.001$, 95% CI: 11.15, 23.90) and perceived informational support with physical and social
23 functioning (B=-6.30, $p=0.05$, 95% CI: -12.52, -0.08; B=8.06, $p=0.02$, 95% CI: 1.17, 14.94). Ideal
24 emotional support was related to physical and social functioning (B=5.80, $p=0.04$, 95% CI: 0.26, 11.34;
25 B=7.53, $p=0.04$, 95% CI: 0.55, 14.51); ideal instrumental support was associated with mental health
26 (B=4.73, $p=0.03$, 95% CI: 0.38, 9.07) and ideal informational support with vitality (B=5.85, $p=0.01$, 95%
27 CI: 1.23, 10.46).
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38 **Conclusion.** Social support was linked to HRQoL in patients with APS. Insufficient social support was
39 associated with limitations in various HRQoL domains. Increasing social support especially through
40 provision of disease-specific education might contribute to improving HRQoL in patients with APS.
41 Patient-tailored interventions addressing psychosocial aspects of living with APS are needed to improve
42 patients' psychological and physical status.
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51 **Key words:** Antiphospholipid (Hughes) syndrome (APS); social support; health-related quality of life
52 (HRQoL); short-form health survey (SF-36)
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1 **Introduction**
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4 Antiphospholipid (Hughes) syndrome (APS) is recognized as a chronic autoimmune disease and is
5 characterized by recurrent venous and arterial thrombosis, miscarriage, neurological features such as
6 stroke, headache, fatigue, memory loss, and epilepsy [1, 2].
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10 Approximately 20% of strokes in people under 40 years and up to 25% of all spontaneous miscarriages
11 (two or more) or fetal losses are due to APS [2, 3]. APS affects predominantly women of childbearing age
12 and is categorized as primary (PAPS) if there is no associated connective tissue disease and secondary
13 (SAPS) if there is, mainly systemic lupus erythematosus (SLE), but also rheumatoid arthritis (RA),
14 Sjogren’s syndrome (SS), scleroderma, vasculitis, and Crohn’s disease [4, 5, 6].
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21 Chronic conditions such RA, SLE, ankylosing spondylitis (AS) and fibromyalgia syndrome (FS) have
22 been shown to have a negative impact on health-related quality of life (HRQoL) especially if there is
23 significant amount of pain involved [4, 5, 7, 8]. Similarly, in APS, HRQoL has been reported to be poorer
24 compared to the general population [9, 10] especially in patients who had a history of arterial thrombosis
25 [10].
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32 The role of the social environment in patients’ HRQoL is very important [11]. The term social support
33 denotes the availability and provision of care and help from an individual’s environment. There are
34 several kinds of social support. Three types of social support often discussed in the literature are
35 distinguished into tangible support such as instrumental support (e.g. assistance with
36 medication/housework), informational support (e.g. education regarding the illness) and treatment
37 strategies and recovery and into intangible support in the form of emotional support (e.g.
38 listening/sympathy) [12]. Social support can be problematic despite people’s good intentions [13] when it
39 involves excessive worry and unsolicited advice, denial of the existence of the illness and its impact on
40 the patient’s life, or support that is not consistent with patients’ beliefs about their condition [13].
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51 Social support is important in improving and maintaining both good physical and mental health in order
52 to self-manage a chronic illness effectively [14, 15]. Support from others, computer-based support and
53 educational interventions combined with self-management [16, 17] and internet support groups [18] have
54 all shown beneficial effects. Elements included in support are also disease-related education such as
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1 diagnosis, treatment, and recovery. Increased levels of disease-specific knowledge were associated with
2 stronger coping skills, perceptions and health behaviours, benefiting disease progression as well as
3 psychological well-being [19, 20]. Social support can also play a mediating role through influencing self-
4 esteem by increasing optimism and decreasing depression thus improving psychological adjustment to
5 chronic illness [21]. Indeed, patients who received more emotional support on a daily basis reported better
6 psychological status than those who did not [22].
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13 Two levels of social support are described: perceived and ideal support. Perceived support refers to the
14 support people perceive to be receiving from their environment while ideal support is the support they
15 would still like to be receiving from friends and family based on their individual needs. It has been argued
16 that the first is more important than the latter because the way patients interpret social support influences
17 psychological adjustment and coping more than ideal support [23, 24]. In addition, lack of understanding
18 from their environment regarding their illness and its consequences contributes to poorer adjustment to
19 chronic illness [23]. In the present study we explored both forms of social support because we attempted
20 to evaluate the magnitude of discrepancy of perceived and ideal social support in patients with APS and
21 whether this discrepancy was associated with their HRQoL. Furthermore, we wanted to assess how much
22 social support patients with APS feel they still need compared to how much they feel they are receiving in
23 order to be able to provide them with additional support to improve their HRQoL.
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37 Availability of social support is related to improved health in patients with rheumatic diseases [25, 26].
38 Particularly, in SLE clinical variables appear to exert a minor influence on patients' HRQoL [5, 27] with
39 psychosocial factors such as social support or helplessness having a significant impact [5, 28]. In
40 addition, "invalidation" referring to lack of understanding or acknowledgment and rejection of the
41 condition has been associated with poorer outcome in patients with rheumatic diseases such as FS and RA
42 [29]. Findings from a literature review on the role of social support in SLE, indicated that social support
43 contributes as a predictor of disease activity, damage and quality of life on both the physical and
44 emotional level [30].
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54 While social support has a beneficial impact on both HRQoL and adjustment to their illness in patients
55 with rheumatic diseases, little is known about the role of social support in HRQoL in patients with APS.
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1 This highlights the need for the present study which aimed to explore the relationship between perceived
2 and ideal social support and HRQoL in APS.
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8 **Methods**

9 **Participant selection and assessment**

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12 This was an internet-based cross-sectional survey. The survey was conducted through a link to an online
13 questionnaire which was available at *KwikSurveys.com*. The link was included in an email that was sent
14 to all members of the Hughes Syndrome Foundation (HSF) worldwide with a request to participate in the
15 survey – if they fulfilled the eligibility criteria. The email was sent by the HSF manager to preserve
16 confidentiality. Participants were given three weeks to complete the survey online after receipt of the
17 email containing the link. As soon as a survey questionnaire was completed, a link leading to each
18 participant’s responses was automatically forwarded to the researchers’ personal email inbox that was set
19 up for the purpose of the study. The link expired 6 months after completion of the survey.
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31 The survey consisted of three sections: (a) the SF-36 assessing health-related quality of life; (b) social
32 support questions; and (c) demographic and disease-specific data. A reminder to complete the survey was
33 sent four days before the expiry of the deadline. The HSF manager forwarded the link to 443 members
34 worldwide to ensure anonymity of the participants. Inclusion criteria were that participants had to be over
35 18 years of age and have a clinical diagnosis of either PAPS or SAPS. The study received approval from
36 the University of Nottingham Research Ethics Committee.
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48 **HRQoL measure**

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51 The SF-36 was employed in the current study due to its generic scope, as well as its reliability and
52 validity in assessing HRQoL in healthy populations [31] and in other diseases [32] despite not having
53 been used in APS before. The SF-36 can also provide data on the influence of the disease on patients’
54 physical psychological and social well-being [33, 34].
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1 Specifically, the SF-36 measures eight domains relative to physical and psychological status which are
2 termed “physical components” (PC) and “mental components” (MC). The physical components include:
3 role physical (RP), general health (GH), bodily pain (BP), and physical functioning (PF) while the mental
4 components encompass: role emotional (RE), vitality (VT), mental health (MH), and social functioning
5 (SF). The self-administered standard version of the SF-36 was selected since it was based on self-
6 completion. Multipoint scales (3 to 10 items) are used to score six of the eight domains while SF and BP
7 are scored on a two-point scale (yes/no). Scale scores are computed by same scale item summation
8 followed by transformation of raw scale score on a range from 0 (lowest possible level of functioning) to
9 100 (highest possible level of functioning) [36].
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20 Demographics and disease-specific information

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23 Demographics included questions on participants’ age, gender and ethnic background. Information on
24 type and time of diagnosis, co-morbidities, date of symptom onset, and number of medications prescribed
25 was collected.
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32 33 **Social support survey**

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36 Social support was assessed on two levels, perceived and ideal, and on three subscales, emotional,
37 instrumental and informational. The survey questions were presented in the form of a set of multiple
38 choice questions with 4 or 5 possible options *displaying various examples of social support scored on a*
39 *two-point scale*. The participants had to indicate which of the available support options listed (e.g. *for*
40 *emotional support: (a) listening; (b) understanding; (c) encouragement; (d) positive feedback; (e)*
41 *willingness to learn more about the illness*) they felt they were receiving (perceived support) and which
42 one(s) they would like to still be receiving (ideal support) by simply ticking the appropriate answer yes or
43 no (please see Appendix 1).
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52 53 54 55 56 **Statistical Analysis**

Participant characteristics summary measures and HRQoL scores were computed as means and standard deviations for continuous (approximate) normally distributed variables and frequencies and percentages for categorical variables. Normality of distribution of continuous summary scales (all p -values >0.05) was assessed with the Shapiro-Wilk test. Multiple linear regression analysis was performed to explore associations between social support and HRQoL in patients in APS and to examine whether perceived and ideal social support were associated with HRQoL. All analyses were adjusted for age. All p -values were two-sided throughout and significance level was set at 5% level. The data were analysed using SPSS version 21.

Results

Participant characteristics

The majority of participants were from the United Kingdom (61.9%). Approximately a quarter were from the United States (24.8%), and fewer from Australia (2.2%), Canada (1.9%) and several other countries. Response rate was 60%. 274 patients completed and returned the questionnaire survey out of a total of 443 individuals who were sent the questionnaire survey link. Four questionnaires were discarded due to insufficient data which resulted in 270 completed questionnaires being included in the analyses. Mean patient age was 45.2 ± 12.1 (range: 18-86 years). The majority of the patients were female (84%; $n=226$) and 45% reported PAPS. Mean age for patients with PAPS was 42.6 ± 11.6 years and for patients with SAPS 47.4 ± 12.1 years. The mean time until receiving a clinical diagnosis for APS was 48.5 ± 87.3 months for PAPS and 75.8 ± 106.4 months for SAPS patients. PAPS patients were prescribed a mean of 3 ± 2.8 medications while SAPS patients 7 ± 5.2 . SLE was reported by 43% ($n=63$) of SAPS participants. On average, participants completed the survey five years post-diagnosis.

Social support

1 Frequency statistics were computed separately for actual and idea social support measures. The majority
2 of patients indicated that they were perceived to be receiving emotional support such as listening (78.7%),
3 understanding (59%), encouragement (55.6%) but not positive feedback (34% vs 66%) and willingness
4 on behalf of their family and friends to learn more about APS (44% vs 56%). In contrast, the majority of
5 patients perceived not to be receiving instrumental support such as help with childcare and
6 housework/shopping (85.1% and 51.9% respectively), provision of transportation (69.8%), financial help
7 (73.5%) and someone to accompany them to GP and hospital appointments (51.5%). Most of the
8 information support, APS patients perceived to be receiving was derived from the internet (82.1%) and
9 support groups (65.7%) whereas a minority of patients reported perceived informational support obtained
10 from GPs (26.9%) and TV or leaflets (14.2%) (Table 1).

11 The main social support (ideal) that patients would like to receive were: understanding (67.9%),
12 willingness to learn more about APS from family or friends (64.9%), information from GPs (74.3%), TV
13 and leaflets (50%) (Table 1).

14 **HRQoL**

15 HRQoL scores were computed for all patients. Mean scores in six domains (RP, BP, GH, VT, SF and RE)
16 were <60 which is the cut-off score reported to indicate highest specificity for functional limitations [51].
17 In the remaining two SF-36 domains (PF and MH), mean scores were >60. The mean HRQoL score was
18 64.4±20.6. Comparison between PAPS and SAPS patients showed poorer HRQoL scores for the PAPS
19 group (< 60) in two domains (GH and VT) and better HRQoL scores (>60) in the remaining six domains
20 (PF, RP, BP, RE, MH and SF) with a mean of 65.15±31.1. For SAPS patients, mean scores were <60 in
21 seven of the SF-36 domains (RP, BP, GH, RE, VT, MH and SF) and >60 in one domain (PF) with a mean
22 of 60.25±23.1.

23 **Relationship between social support and HRQoL in APS**

24 **Perceived social support and HRQoL**

1 Perceived social support was assessed on three levels: emotional, instrumental and informational. On the
2
3 perceived emotional level, results showed that higher levels of encouragement were related to better
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5 physical functioning (B=7.77, p<0.01; 95%CI: 2.25, 13.29), better role physical functioning (B=15.83;
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7 p<0.01; 95%CI: 3.96, 27.70) and better general health (B=5.62; p<0.01; 95%CI: 1.02, 10.22) while less
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9 understanding from friends and family were associated with lower levels of vitality (B=-6.22, p<0.01;
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11 95%CI:-10.91, -1.53). Lower provision of positive feedback was associated with poorer role emotional
12
13 functioning (B=-13.88, p<0.05; 95%CI: -26.51, -1.25) and poorer mental health (B=-7.99, p<0.001;
14
15 95%CI: -12.52, -3.47) and similarly lower degree of willingness to learn more about APS was also related
16
17 to poorer mental health (B=-4.27, p<0.05; 95%CI: -8.51, -0.02) (Table 2).

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20 Results on perceived instrumental support indicated that lower provision of help with childcare was
21
22 related to more limited social functioning (B=-9.21, p<0.05; 95%CI: -18.49, 0.07) whereas more support
23
24 in terms of helping with housework and/or shopping were associated with better physical functioning
25
26 (B=13.50, p<0.001; 95%CI: 8.17, 18.84), role physical (B=18.64, p<0.01; 95%CI: 6.88, 30.40), lower
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28 bodily pain (B=17.52, p<0.001; 95%CI: 11.15, 23.90), better general health (B=10.40, p<0.001; 95%CI:
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30 5.94, 14.87), higher levels of vitality (B=8.85, p<0.001; 95%CI: 4.30, 13.39), and better social
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32 functioning (B=9.22, p<0.01; 95%CI: 2.71, 15.73). Provision of transportation was associated with better
33
34 HRQoL in all domains except for mental health while financial help was associated with better physical
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36 functioning (B=7.93, p<0.01; 95%CI: 1.70, 14.16) and lower bodily pain (B=9.31, p<0.05; 95%CI: 1.77,
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38 16.85). Attendance at General Practitioner (GP) and hospital appointments was also related to better
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40 HRQoL in the domains physical functioning (B=9.20, p<0.001; 95%CI: 3.71, 14.69), bodily pain
41
42 (B=9.74, p<0.01; 95%CI: 3.09, 16.39), general health (B=5.53, p<0.05; 95%CI: 0.92, 10.14), and role
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44 emotional (B=12.14, p<0.05; 95%CI: 0.12, 24.15) (see Table 3).

45
46
47 Perceived informational support was associated with HRQoL in terms of information provided by GPs,
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49 support groups, and consultants/charity. Less information provided by GPs was associated with more
50
51 limited physical functioning (B=-6.30, p<0.05; 95%CI: -12.52, -0.08), and poorer role physical
52
53 performance (B=-19.37, p<0.01; 95%CI: -32.63, -6.11), more support provided by support groups was
54
55 related to better social functioning (B=8.06, p<0.05; 95%CI: 1.17, 14.94) and less information from
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1 consultants/charity was related to poorer general health (B=-8.67, p<0.05; 95%CI: -16.72, -0.61) (Table
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9 **Ideal social support and HRQoL**

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11 Ideal social support was also assessed on three levels: emotional, instrumental and informational. Patients
12 reported the levels of social support they felt they still wanted to receive based on their needs. People who
13 felt they still needed someone to listen to their concerns and worries would be more likely to have better
14 physical functioning if they had this support (B=5.80, p<0.05; 95%CI: 0.26, 11.34) and higher levels of
15 vitality (B=6.91, p<0.01; 95%CI: 2.32, 4.51). The need for understanding was linked to better HRQoL
16 except for bodily pain and role emotional and the need for more frequent encouragement was associated
17 with better physical functioning (B=7.78, p<0.01; 95%CI: 2.30, 13.26), role physical (B=14.46, p<0.05;
18 95%CI: 2.66, 26.26), and greater vitality (B=5.16, p<0.05; 95%CI: 0.56, 9.77). Greater provision of
19 positive feedback was associated with better role physical (B=16.35, p<0.01; 95%CI: 4.59, 28.12), greater
20 vitality (B=5.29, p<0.05; 95%CI: 0.69, 9.90), and role emotional (B=13.13, p<0.05; 95%CI: 1.23, 25.04).
21 Patients' greater need for their family and friends to learn more about APS was related to better role
22 physical (B=15.99, p<0.01; 95%CI: 3.62, 28.36), and greater vitality (B=7.21, p<0.01; 95%CI: 2.42,
23 12.01) (Table 5).
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39 Ideal instrumental support was associated with better HRQoL in all domains in terms of provision of help
40 with housework and/or shopping and attendance at GP and hospital appointments (see Table 6). The need
41 for provision of transportation was related to better HRQoL in all domains except role physical and role
42 emotional (see Table 6).
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48 There was an association between greater informational support provided by and better role emotional
49 (B=19.05, p<0.01; 95%CI: 5.39, 32.70) and better mental health (B=6.29, p<0.01; 95%CI: 1.35, 11.23),
50 while information from support groups was associated with greater vitality (B=5.85, p<0.01; 95%CI:
51 1.23, 10.46) (Table 7).
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Discrepancy values between perceived and ideal social support

In order to examine discrepancy between actual and ideal social support in APS patients, the total values of each were computed for all three types of social support, i.e. emotional, instrumental and informational. Following that, a discrepancy value was obtained by subtracting the total actual support values from the total ideal support values for each of the three types of social support. The results indicated that the mean of the discrepancy value between actual and ideal emotional support ($M=.064$; $SD=2.42$; $N=268$) as well as the mean of the discrepancy between actual and ideal instrumental support ($M=-.011$; $SD=1.24$; $N=268$) were relatively small. In contrast, the mean of the discrepancy between ideal and actual informational support ($M=.23$; $SD=1.58$; $N=268$) was large. Thus, it would seem that the informational support APS patients perceive to be receiving is far smaller than the support they ideally would need to be receiving. As far as informational and instrumental support was concerned, the ideal levels of social support did not appear to differ significantly from the perceived levels.

Discussion

This is the first study to examine associations between social support and HRQoL in patients with APS. HRQoL in individuals living with rheumatic and autoimmune diseases is affected to a significant extent [4, 5] with social support playing a significant role [18, 37-39]. However, very little research has been conducted into HRQoL and social support in patients with APS so far, to our knowledge. A variety of measures are available to assess HRQoL including generic and disease-specific instruments. Generic instruments can be employed across a range of different conditions while specific instruments are disease-adapted. APS-related symptoms vary significantly and can have an impact on patients' physical, social and emotional status [1].

Associations between perceived social support and HRQoL indicated that higher provision of instrumental support was related to better HRQoL in terms of patients' physical and mental status. Higher perceived emotional support, on the other hand, led to better mental health status probably because increased support in this domain may contribute to patients getting more time to rest, socialize and experience less stress [43-46]. More perceived information provided by GPs was also related to better

1 physical functioning and role physical, while information received from support groups was related to
2 better social functioning and less information from consultants and the charity were related to poorer
3 general health. This is in accordance with previous literature which suggests that doctor-patient
4 communication plays a significant role in patients' health outcomes, quality of life, absence from work
5 and treatment adherence [47]. It could be assumed that because of insufficient information provided,
6 patients are not aware of the degree they should engage in various activities and this subsequently may
7 have a negative effect on their physical health due to increased or decreased involvement.
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16 Similarly, higher ideal social support was also linked to better HRQoL. People who felt they needed
17 someone to listen to their concerns and worries were predicted to be more likely to have better physical
18 functioning such as fewer limitations in performing physical activities, including bathing or dressing and
19 experience greater vitality i.e. feeling more energetic. This could be attributed to the fact that people
20 might be likely to receive some reassurance from their social circle that they are able to perform certain
21 activities following the expression of their concerns or offer them additional support which might
22 contribute to decrease the burden that affect their energy levels. This was also reflect in the fact that the
23 need for more frequent encouragement was associated with better physical while greater provision of
24 positive feedback was associated with better role physical greater vitality, and role emotional.
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35 The need to feel understood by family and friends was linked to better HRQoL in all domains except for
36 physical pain and role emotional. Indeed, negative social responses, particularly discounting (rejecting)
37 and lack of understanding (not being acknowledged), were associated with poorer health among patients
38 with fibromyalgia and rheumatoid arthritis [52]. Greater understanding would be derived from better
39 knowledge of APS by the patient's family and social circle. Results showed that patients' greater need for
40 their family and friends to learn more about APS was related to better role physical, and greater vitality.
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1 for emotional and instrumental support. However, there was a statistically significant difference between
2 the informational support patients reported to be receiving and the informational support they felt they
3 were still lacking highlighting the lack of awareness as well as health professional education and public
4 awareness about APS. More knowledge of APS provided by health care professionals and the media
5 might enhance patients' coping with the disease due to increased education on aspects such as self-
6 management, and medication and more effective and helpful support from their families and friends.
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14 There are some limitations to this study. First, the participants were members of the HSF which might
15 have increased the likelihood of receiving higher social support compared to patients who do not belong
16 to a charity. The data was based on self-report thus environmental or emotional influences could not be
17 controlled. Diagnosis of APS could not be confirmed due to lack of access to patients' medical records or
18 physical and laboratory examination. The survey was cross-sectional which prevents detection of change
19 over time and assessment of causal relationships. Findings cannot be generalized due to the fact that the
20 majority of patients were female and British. Ethnicity and culture has been suggested to affect perceived
21 quality of life of individuals on dialysis after renal transplant with Asian renal patients perceiving HRQoL
22 more negatively than white Europeans [47]. Factors such as major life events, for example death, divorce
23 or severity of disease and depression status were not controlled for, any of which could potentially be
24 related to poorer HRQoL in either group. Many factors such as bereavement, financial difficulties,
25 depression and anxiety were also found to place a burden on HRQoL [11].
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39 Strengths of this study, on the other hand, include a high response rate (60%), a satisfactory sample size
40 providing sufficient power for analyses. The fact that a relatively non-researched patient population was
41 assessed was a further advantage. Examination of all aspects of patients' well-being relating to the
42 psychological, social and physical impact of APS and its influence by social support measures provided a
43 more holistic approach and increased understanding of the degree and nature of the disease impact on
44 patients' HRQoL.
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52 Findings indicated that certain types of social support exert a significant influence on a variety of domains
53 in APS patients' HRQoL. Patients reported receiving insufficient social support. By extension, this might
54 be suggestive of the beneficial effect of social support on HRQoL in patients with APS either through
55 medication adherence or also through more effective coping skills. Lack of support in terms of providing
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1 disease – and medication-specific information has been associated with decreased medication adherence
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3 in patients with autoimmune diseases [48]. In addition, increased informational support especially by
4
5 knowledgeable health professionals might improve provision of support by patients’ family and friends
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7 through reducing “invalidation” [29]. Particularly, due to the multi-faceted nature of APS, as is true of
8
9 most autoimmune diseases, involving pain, disability, uncertainty about its progression and fear of
10
11 treatment effects and based on the current findings it can be suggested that a combination of approaches
12
13 and interventions could prove to be of great importance and help in improving adjustment and coping
14
15 with APS. This combination would need to incorporate social approach and support from practitioners,
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17 family, friends, and co-workers, as well as elements from both the bio-psychosocial and biomedical
18
19 frameworks [49, 50] tailored to the needs of APS patients. Specific strategies could be implemented
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21 through both primary and secondary care and include patient- and family/friends-education sessions
22
23 delivered by specialist nurses. These sessions could provide disease- and treatment-specific information
24
25 and self-management strategies such as International Normalised Ratio (INR) measuring, dietary advice
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27 and pacing to patients as well as disease-related information and ways of supporting their loved one in
28
29 coping more effectively with APS to families/friends of patients with APS.
30

31 **Acknowledgements**

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38 This research was supported by the Institute of Work, Health & Organisations of the University of
39
40 Nottingham. We are grateful to Ms. Kate Hindle, manager of the Hughes Syndrome Foundation, for her
41
42 invaluable help and support in conducting the study as well as to the Hughes Syndrome London support
43
44 group for their comments and helpful suggestions. We would also like to acknowledge the significant
45
46 contribution of all the Hughes Syndrome Foundation members who participated in the present study. Last
47
48 but not least, we would like to express our gratitude to Professor Graham Hughes for his encouragement,
49
50 support and passion which inspired this research and motivated us throughout the whole project. The
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52 views expressed here are the authors’ and do not necessarily reflect those of any other person or
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54 organization.
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1 **Funding**
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4 This research ideal no specific grant from any funding agency in the public, commercial, or not-for-profit
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6 sectors.
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11 **Conflict of interest**
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Table 1 Computed values of results for ideal and actual social support

| | Actual Social Support - Emotional (n=268) | | | Ideal Social Support - Emotional (n=268) | |
|--|--|-------------|--|---|-------------|
| | Yes | No | | Yes | No |
| Listening | 211 (78.7%) | 57 (21.3%) | Listening | 136 (50.7%) | 132 (49.3%) |
| Understanding | 158 (59%) | 110 (41%) | Understanding | 182 (67.9%) | 86 (32.1%) |
| Encouragement | 149 (55.6%) | 119 (44.4%) | Encouragement | 127 (47.4%) | 141 (52.6%) |
| Positive Feedback | 91 (34%) | 177 (66%) | Positive Feedback | 125 (46.6%) | 143 (53.4%) |
| Willingness to learn more about APS | 118 (44%) | 150 (56%) | Willingness to learn more about APS | 174 (64.9%) | 94 (35.1%) |
| | Actual Social Support - Instrumental (n=268) | | | Ideal Social Support - Instrumental (n=268) | |
| Help with childcare | 40 (14.9) | 228 (85.1%) | Help with childcare | 41 (15.3%) | 227 (84.7%) |
| Help with housework/ shopping | 129 (48.1%) | 139 (51.9%) | Help with housework/ shopping | 121 (45.1%) | 147 (54.9%) |
| Provision of transportation | 81 (30.2%) | 187 (69.8%) | Provision of transportation | 66 (24.6%) | 202 (75.4%) |
| Financial help | 71 (26.5%) | 197 (73.5%) | Financial help | 68 (25.4%) | 200 (74.6%) |
| Attendance GPs/hospital appt | 131 (48.9%) | 137 (51.5%) | Attendance GPs/hospital appt | 109 (40.7%) | 159 (59.3%) |
| | Actual Social Support - Informational (n=268) | | | Ideal Social Support - Informational (n=268) | |
| Information provided by GPs | 72 (26.9%) | 196 (73.1%) | Information provided by GPs | 199 (74.3%) | 69 (25.7%) |
| Information provided on the internet | 220 (82.1%) | 48 (17.9%) | Information provided on the internet | 119 (44.4%) | 149 (55.6%) |
| Information provided by support groups | 176 (65.7%) | 92 (34.3%) | Information provided by support groups | 119 (44.4%) | 149 (55.6%) |
| Information provided on TV/ leaflets | 38 (14.2%) | 229 (85.4%) | Information provided on TV/ leaflets | 134 (50.0%) | 134 (50.0%) |
| Information provided by consultants/charity (n=20) | 19 (7.1%) | 1 (0.4%) | Information provided by consultants/charity (n=42) | 41 (15.2%) | 1 (0.4%) |

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Table 2 Associations between perceived emotional support and HRQoL in APS

| <i>Perceived Social Support – Emotional (n= 270)</i> | | | | | |
|--|-------------------------|----------------------------|--------------------------|-----------------------------|--|
| <i>SF-36 domains</i> | <i>Listening</i> | <i>Understanding</i> | <i>Encouragement</i> | <i>Positive Feedback</i> | <i>Willingness to learn more about APS</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | -0.50 (-7.29, 6.30) | -2.62 (-8.31, 3.06) | 7.77** (2.25, 13.29) | 0.43 (-5.47, 6.33) | 4.21 (-1.21, 9.62) |
| Role physical | 5.90 (-8.68, 20.48) | -5.60 (-17.79, 6.60) | 15.83** (3.96, 27.70) | 4.84 (-7.82, 17.50) | 0.32 (-11.37, 12.01) |
| Bodily pain | 2.97 (-5.25, 11.19) | -2.23 (-9.08, 4.62) | 6.62 (-0.08, 13.33) | 0.43 (-6.68, 7.53) | 1.35 (-5.21, 7.91) |
| General health | -2.74 (-8.40, 2.92) | -1.74 (-6.46, 2.98) | 5.62** (1.02, 10.22) | -0.45 (-5.35, 4.46) | 0.57 (-3.96, 5.10) |
| <i>MC</i> | | | | | |
| Vitality | -3.66 (-9.35, 2.03) | -6.22** (-10.91, -1.53) | 1.74 (-2.94, 6.41) | -4.23 (-9.13, 0.68) | -3.62 (-8.15, 0.92) |
| Social functioning | -3.82 (-11.88, 4.25) | -4.60 (-11.30, 2.11) | 1.82 (-4.80, 8.43) | -3.64 (-10.61, 3.33) | -3.03 (-9.47, 3.41) |
| Role emotional | -9.60 (-24.24, 5.03) | -7.56 (-19.83, 4.71) | -2.45 (-14.55, 9.65) | -13.88* (-26.51, -1.25) | -10.38 (-22.06, 1.31) |
| Mental health | -3.44 (-8.78, 1.90) | -3.99 (-8.43, 0.44) | -2.57 (-6.95, 1.81) | -7.99*** (-12.52, -3.47) | -4.27* (-8.51, -0.02) |

Multiple Linear Analysis examining the association between perceived emotional support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 3 Associations between perceived instrumental support and HRQoL in APS

| <i>Perceived Social Support – Instrumental (n= 270)</i> | | | | | |
|---|----------------------------|--------------------------------------|------------------------------------|-------------------------|---|
| <i>SF-36 domains</i> | <i>Help with childcare</i> | <i>Help with housework/ shopping</i> | <i>Provision of transportation</i> | <i>Financial help</i> | <i>Attendance at GPs/ hospital appointments</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | 0.24 (-7.63, 8.10) | 13.50*** (8.17, 18.84) | 15.03*** (9.26, 20.81) | 7.93** (1.70, 14.16) | 9.20*** (3.71, 14.69) |
| Role physical | 1.09 (-15.83, 17.99) | 18.64** (6.88, 30.40) | 20.20** (7.40, 32.99) | 11.21 (-2.28, 26.69) | 8.92 (-3.06, 20.91) |
| Bodily pain | 0.15 (-9.50, 9.81) | 17.52*** (11.15, 23.90) | 14.59*** (7.51, 21.67) | 9.31* (1.77, 16.85) | 9.74** (3.09, 16.39) |
| General health | 0.75 (-5.82, 7.31) | 10.40*** (5.94, 14.87) | 6.03* (1.03, 11.03) | 3.88 (-1.36, 9.12) | 5.53* (0.92, 10.14) |
| <i>MC</i> | | | | | |
| Vitality | -0.08 (-6.69, 6.52) | 8.85*** (4.30, 13.39) | 8.48*** (3.50, 13.46) | 1.67 (-3.62, 6.95) | 1.18 (-3.50, 5.87) |
| Social functioning | -9.21* (-18.49, 0.07) | 9.22** (2.71, 15.73) | 10.43** (3.34, 17.52) | 2.10 (-5.39, 9.58) | 5.26 (-1.34, 11.86) |
| Role emotional | -8.29 (-25.25, 8.67) | 5.59 (-6.45, 17.62) | 14.95* (1.97, 27.92) | 7.15 (-6.44, 20.74) | 12.14* (0.12, 24.15) |
| Mental health | -3.26 (-9.44, 2.93) | 2.46 (-1.91, 6.83) | 3.63 (-1.12, 8.38) | 0.33 (-4.63, 5.30) | -0.22 (-4.62, 4.18) |

Multiple Linear Analysis examining the association between perceived instrumental support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 4 Associations between perceived informational support and HRQoL in APS

| <i>Perceived Social Support – Informational (n= 270)</i> | | | | | |
|--|------------------------------------|---|---|--|--|
| <i>SF-36 domains</i> | <i>Information provided by GPs</i> | <i>Information provided on the internet</i> | <i>Information provided by support groups</i> | <i>Information provided on TV/leaflets</i> | <i>Information provided by consultants/charity</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | -6.30* (-12.52, -0.08) | -2.17 (-9.50, 5.16) | 4.20 (-1.65, 10.05) | 4.42 (-3.49, 12.33) | -3.02 (-12.74, 6.69) |
| Role physical | -19.37** (-32.63, -6.11) | -3.52 (-19.16, 12.11) | 6.93 (-5.65, 19.51) | 8.03 (-8.99, 25.05) | -14.63 (-35.45, 6.20) |
| Bodily pain | -6.12 (-13.63, 1.39) | 3.02 (-5.65, 11.69) | 4.57 (-2.50, 11.64) | 6.15 (-3.37, 15.66) | -5.52 (-17.19, 6.15) |
| General health | -0.50 (-5.71, 4.70) | 0.41 (-5.61, 6.43) | 2.07 (-2.80, 6.95) | 2.89 (-3.66, 9.44) | -8.67* (-16.72, -0.61) |
| <i>MC</i> | | | | | |
| Vitality | -4.59 (-9.80, 0.62) | 1.50 (-4.56, 7.56) | 0.65 (-4.26, 5.56) | -2.40 (-8.99, 4.19) | -1.18 (-9.35, 6.99) |
| Social functioning | -6.79 (-14.16, 0.58) | -2.63 (-11.20, 5.94) | 8.06* (1.17, 14.94) | 8.86 (-0.42, 18.14) | -9.34 (-20.85, 2.18) |
| Role emotional | -11.02 (-24.49, 2.40) | -13.45 (-29.21, 2.31) | 5.10 (-7.57, 17.76) | -2.99 (-20.12, 14.13) | -8.62 (-29.59, 12.36) |
| Mental health | -1.12 (-6.03, 3.80) | -4.08 (-9.74, 1.59) | -1.09 (-5.69, 3.52) | -1.71 (-7.90, 4.48) | -3.72 (-11.38, 3.94) |

Multiple Linear Analysis examining the association between perceived informational support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 5 Associations between ideal emotional support and HRQoL in APS

| <i>Ideal Social Support – Emotional (n= 270)</i> | | | | | |
|--|------------------------|--------------------------|-------------------------|--------------------------|--|
| <i>SF-36 domains</i> | <i>Listening</i> | <i>Understanding</i> | <i>Encouragement</i> | <i>Positive Feedback</i> | <i>Willingness to learn more about APS</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | 5.80* (0.26, 11.34) | 6.21* (0.31, 12.10) | 7.78** (2.30, 13.26) | 3.99 (-1.54, 9.54) | 4.91 (-0.89, 10.71) |
| Role physical | 5.76 (-6.21, 17.73) | 14.27* (1.61, 26.93) | 14.46* (2.66, 26.26) | 16.35** (4.59, 28.12) | 15.99** (3.62, 28.36) |
| Bodily pain | 0.23 (-6.50, 6.96) | 5.87 (-1.26, 12.99) | 2.66 (-4.04, 9.35) | -0.005 (-6.70, 6.69) | 0.75 (-6.27, 7.76) |
| General health | 2.06 (-2.57, 6.70) | 5.66* (0.77, 10.56) | 3.64 (-0.96, 8.24) | 1.41 (-3.21, 6.02) | 2.30 (-2.53, 7.14) |
| <i>MC</i> | | | | | |
| Vitality | 6.91** (2.32, 4.51) | 9.48*** (4.64, 14.32) | 5.16* (0.56, 9.77) | 5.29* (0.69, 9.90) | 7.21** (2.42, 12.01) |
| Social functioning | 5.55 (-1.03, 12.12) | 7.53* (0.55, 14.51) | 6.31 (-0.23, 12.85) | 4.10 (-2.47, 10.66) | 4.77 (-2.10, 11.65) |
| Role emotional | 9.91 (-2.10, 21.92) | 4.88 (-7.95, 17.72) | 7.69 (-4.29, 19.67) | 13.13* (1.23, 25.04) | 8.41 (-4.15, 20.96) |
| Mental health | 2.32 (-2.06, 6.69) | 6.17** (1.57, 10.78) | 2.55 (-1.81, 6.90) | 1.68 (-2.68, 6.04) | 1.74 (-2.83, 6.30) |

Multiple Linear Analysis examining the association between ideal emotional support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 6 Associations between ideal instrumental support and HRQoL in APS

| <i>Ideal Social Support – Instrumental (n= 270)</i> | | | | | |
|---|----------------------------|--------------------------------------|------------------------------------|-------------------------|---|
| <i>SF-36 domains</i> | <i>Help with childcare</i> | <i>Help with housework/ shopping</i> | <i>Provision of transportation</i> | <i>Financial help</i> | <i>Attendance at GPs/ hospital appointments</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | -0.64 (-8.56, 7.28) | 10.77*** (5.33, 16.20) | 11.28*** (4.99, 17.58) | 3.67 (-2.72, 10.07) | 11.81*** (6.29, 17.33) |
| Role physical | -1.50 (-18.54, 15.54) | 20.03*** (8.29, 31.77) | 12.93 (-0.83, 26.69) | 11.59 (-2.05, 25.24) | 18.78** (6.77, 30.79) |
| Bodily pain | -4.52 (-14.13, 5.09) | 12.01*** (5.45, 18.58) | 10.63** (2.95, 18.31) | 4.50 (-3.16, 12.17) | 9.87** (3.12, 16.62) |
| General health | 0.75 (-5.82, 7.31) | 8.48*** (3.95, 13.00) | 5.58* (0.25, 10.91) | 2.41 (-2.91, 7.73) | 7.36** (2.71, 12.01) |
| <i>MC</i> | | | | | |
| Vitality | 3.91 (-2.68, 10.50) | 11.69*** (7.24, 16.14) | 9.35*** (4.07, 14.64) | 5.17 (-0.15, 10.49) | 9.86*** (5.25, 14.47) |
| Social functioning | 0.83 (-8.52, 10.18) | 11.82*** (5.37, 18.28) | 11.88*** (4.37, 19.40) | 1.64 (-5.94, 9.22) | 9.94** (3.31, 16.58) |
| Role emotional | 0.97 (-16.15, 18.08) | 16.75** (4.85, 28.66) | 12.12 (-1.72, 25.96) | 10.53 (-3.26, 24.32) | 14.28* (2.07, 26.48) |
| Mental health | 0.83 (-5.37, 7.03) | 4.73* (0.38, 9.07) | 5.79* (0.77, 10.82) | 3.56 (-1.45, 8.57) | 4.46* (0.02, 8.90) |

Multiple Linear Analysis examining the association between ideal instrumental support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 7 Associations between ideal informational support and HRQoL in APS

| <i>Ideal Social Support – Informational (n= 270)</i> | | | | | |
|--|------------------------------------|---|---|--|--|
| <i>SF-36 domains</i> | <i>Information provided by GPs</i> | <i>Information provided on the internet</i> | <i>Information provided by support groups</i> | <i>Information provided on TV/leaflets</i> | <i>Information provided by consultants/charity</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | 2.23 (-4.18, 8.64) | -0.64 (-6.27, 4.99) | 4.18 (-1.39, 9.75) | 3.20 (-2.37, 8.76) | -4.12 (-8.82, 0.59) |
| Role physical | -0.63 (-14.40, 13.15) | -1.76 (-13.84, 10.32) | 6.04 (-5.94, 18.01) | 11.41 (-0.48, 23.30) | 1.37 (-8.80, 11.53) |
| Bodily pain | 1.99 (-5.68, 9.66) | -1.46 (-8.25, 5.32) | 2.19 (-4.55, 8.92) | 5.00 (-1.69, 11.69) | -0.77 (-6.62, 5.07) |
| General health | 0.42 (-4.87, 5.71) | 1.75 (-2.91, 6.42) | 2.68 (-1.95, 7.31) | 1.01 (-3.63, 5.64) | -0.79 (-4.74, 3.16) |
| <i>MC</i> | | | | | |
| Vitality | 3.34 (-1.97, 8.65) | 4.24 (-0.44, 8.91) | 5.85** (1.23, 10.46) | 2.40 (-2.25, 7.06) | -1.33 (-5.30, 2.65) |
| Social functioning | 2.29 (-5.24, 9.82) | 0.20 (-6.46, 6.85) | 3.18 (-3.43, 9.78) | 1.69 (-4.91, 8.29) | -1.93 (-7.55, 3.70) |
| Role emotional | 19.05** (5.39, 32.70) | -2.36 (-14.52, 9.81) | 7.70 (-4.34, 19.75) | 11.27 (-0.70, 23.25) | -2.34 (-12.55, 7.87) |
| Mental health | 6.29** (1.35, 11.23) | 3.49 (-0.91, 7.88) | 3.81 (-0.55, 8.17) | 2.19 (-2.18, 6.55) | -1.01 (-4.74, 2.72) |

Multiple Linear Analysis examining the association between ideal informational support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Modern Rheumatology

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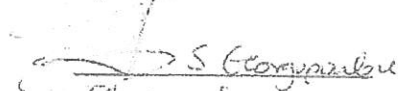
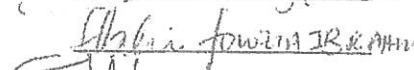
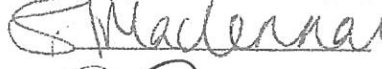
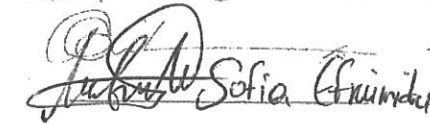
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1 (B=-6.30, $p=0.05$, 95% CI: -12.52, -0.08; B=8.06, $p=0.02$, 95% CI: 1.17, 14.94). Ideal
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3 emotional support was related to physical and social functioning (B=5.80, $p=0.04$, 95%
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5 CI: 0.26, 11.34; B=7.53, $p=0.04$, 95% CI: 0.55, 14.51); ideal instrumental support was
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7 associated with mental health (B=4.73, $p=0.03$, 95% CI: 0.38, 9.07) and ideal
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9 informational support with vitality (B=5.85, $p=0.01$, 95% CI: 1.23, 10.46).
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12
13 **Conclusion.** Social support was linked to HRQoL in patients with APS. Insufficient
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15 social support was associated with limitations in various HRQoL domains. Increasing
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17 social support especially through provision of disease-specific education might
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19 contribute to improving HRQoL in patients with APS. Patient-tailored interventions
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21 addressing psychosocial aspects of living with APS are needed to improve patients'
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23 psychological and physical status.
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32 **Key words:** Antiphospholipid (Hughes) syndrome (APS); social support; health-related
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34 quality of life (HRQoL); short-form health survey (SF-36)
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Introduction

Antiphospholipid (Hughes) syndrome (APS) is recognized as a chronic autoimmune disease and is characterized by recurrent venous and arterial thrombosis, miscarriage, neurological features such as stroke, headache, fatigue, memory loss, and epilepsy [1, 2].

Approximately 20% of strokes in people under 40 years and up to 25% of all spontaneous miscarriages (two or more) or fetal losses are due to APS [2, 3]. APS affects predominantly women of childbearing age and is categorized as primary (PAPS) if there is no associated connective tissue disease and secondary (SAPS) if there is, mainly systemic lupus erythematosus (SLE), but also rheumatoid arthritis (RA), Sjogren's syndrome (SS), scleroderma, vasculitis, and Crohn's disease [4, 5, 6].

Chronic conditions such RA, SLE, ankylosing spondylitis (AS) and fibromyalgia syndrome (FS) have been shown to have a negative impact on health-related quality of life (HRQoL) especially if there is significant amount of pain involved [4, 5, 7, 8]. Similarly, in APS, HRQoL has been reported to be poorer compared to the general population [9, 10] especially in patients who had a history of arterial thrombosis [10].

The role of the social environment in patients' HRQoL is very important [11]. The term social support denotes the availability and provision of care and help from an individual's environment. There are several kinds of social support. Three types of social support often discussed in the literature are distinguished into tangible support such as instrumental support (e.g. assistance with medication/housework), informational support (e.g. education regarding the illness) and treatment strategies and recovery and

1 into intangible support in the form of emotional support (e.g. listening/sympathy) [12].
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3 Social support can be problematic despite people's good intentions [13] when it
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5 involves excessive worry and unsolicited advice, denial of the existence of the illness
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7 and its impact on the patient's life, or support that is not consistent with patients' beliefs
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9 about their condition [13].
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13 Social support is important in improving and maintaining both good physical and
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15 mental health in order to self-manage a chronic illness effectively [14, 15]. Support
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17 from others, computer-based support and educational interventions combined with self-
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19 management [16, 17] and internet support groups [18] have all shown beneficial effects.
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21 Elements included in support are also disease-related education such as diagnosis,
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23 treatment, and recovery. Increased levels of disease-specific knowledge were associated
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25 with stronger coping skills, perceptions and health behaviours, benefiting disease
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27 progression as well as psychological well-being [19, 20]. Social support can also play a
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29 mediating role through influencing self-esteem by increasing optimism and decreasing
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31 depression thus improving psychological adjustment to chronic illness [21]. Indeed,
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33 patients who received more emotional support on a daily basis reported better
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35 psychological status than those who did not [22].
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44 Two levels of social support are described: perceived and ideal support. Perceived
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46 support refers to the support people perceive to be receiving from their environment
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48 while ideal support is the support they would still like to be receiving from friends and
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50 family based on their individual needs. It has been argued that the first is more
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52 important than the latter because the way patients interpret social support influences
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54 psychological adjustment and coping more than ideal support [23, 24]. In addition, lack
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1 of understanding from their environment regarding their illness and its consequences
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3 contributes to poorer adjustment to chronic illness [23]. In the present study we
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5 explored both forms of social support because we attempted to evaluate the magnitude
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7 of discrepancy of perceived and ideal social support in patients with APS and whether
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9 this discrepancy was associated with their HRQoL. Furthermore, we wanted to assess
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11 how much social support patients with APS feel they still need compared to how much
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13 they feel they are receiving in order to be able to provide them with additional support
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15 to improve their HRQoL.
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21 Availability of social support is related to improved health in patients with rheumatic
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23 diseases [25, 26]. Particularly, in SLE clinical variables appear to exert a minor
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25 influence on patients' HRQoL [5, 27] with psychosocial factors such as social support
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27 or helplessness having a significant impact [5, 28]. In addition, "invalidation" referring
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29 to lack of understanding or acknowledgment and rejection of the condition has been
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31 associated with poorer outcome in patients with rheumatic diseases such as FS and RA
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33 [29]. Findings from a literature review on the role of social support in SLE, indicated
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35 that social support contributes as a predictor of disease activity, damage and quality of
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37 life on both the physical and emotional level [30].
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44 While social support has a beneficial impact on both HRQoL and adjustment to their
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46 illness in patients with rheumatic diseases, little is known about the role of social
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48 support in HRQoL in patients with APS. This highlights the need for the present study
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50 which aimed to explore the relationship between perceived and ideal social support and
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52 HRQoL in APS.
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Methods

Participant selection and assessment

This was an internet-based cross-sectional survey. The survey was conducted through a link to an online questionnaire which was available at *KwikSurveys.com*. The link was included in an email that was sent to all members of the **Hughes Syndrome Foundation (HSF)** worldwide with a request to participate in the survey – if they fulfilled the eligibility criteria. The email was sent by the **HSF** manager to preserve confidentiality. Participants were given three weeks to complete the survey online after receipt of the email containing the link. As soon as a survey questionnaire was completed, a link leading to each participant's responses was automatically forwarded to the researchers' personal email inbox that was set up for the purpose of the study. The link expired 6 months after completion of the survey.

The survey consisted of three sections: (a) the SF-36 assessing health-related quality of life; (b) social support questions; and (c) demographic and disease-specific data. A reminder to complete the survey was sent four days before the expiry of the deadline. The **HSF** manager forwarded the link to 443 members worldwide to ensure anonymity of the participants. Inclusion criteria were that participants had to be over 18 years of age and have a clinical diagnosis of either **PAPS or SAPS**. The study received approval from the University of Nottingham Research Ethics Committee.

HRQoL measure

The SF-36 was employed in the current study due to its generic scope, as well as its reliability and validity in assessing **HRQoL** in healthy populations [31] and in other

1 diseases [32] despite not having been used in APS before. The SF-36 can also provide
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3 data on the influence of the disease on patients' physical psychological and social well-
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5 being [33, 34].
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9 Specifically, the SF-36 measures eight domains relative to physical and psychological
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11 status which are termed "physical components" (PC) and "mental components" (MC).
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13 The physical components include: role physical (RP), general health (GH), bodily pain
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15 (BP), and physical functioning (PF) while the mental components encompass: role
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17 emotional (RE), vitality (VT), mental health (MH), and social functioning (SF). The
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19 self-administered standard version of the SF-36 was selected since it was based on self-
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21 completion. Multipoint scales (3 to 10 items) are used to score six of the eight domains
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23 while SF and BP are scored on a two-point scale (yes/no). Scale scores are computed by
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25 same scale item summation followed by transformation of raw scale score on a range
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27 from 0 (lowest possible level of functioning) to 100 (highest possible level of
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29 functioning) [36].
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37 Demographics and disease-specific information

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39 Demographics included questions on participants' age, gender and ethnic background.
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41 Information on type and time of diagnosis, co-morbidities, date of symptom onset, and
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43 number of medications prescribed was collected.
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48 Social support survey

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51 Social support was assessed on two levels, perceived and ideal, and on three subscales,
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53 emotional, instrumental and informational. The survey questions were presented in the
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1 form of a set of multiple choice questions with 4 or 5 possible options displaying
2
3 various examples of social support scored on a two-point scale. The participants had to
4
5 indicate which of the available support options listed (e.g. for emotional support: (a)
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7 listening; (b) understanding; (c) encouragement; (d) positive feedback; (e) willingness
8
9 to learn more about the illness) they felt they were receiving (perceived support) and
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11 which one(s) they would like to still be receiving (ideal support) by simply ticking the
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13 appropriate answer yes or no (please see Appendix 1).
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19 Statistical Analysis

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22 Participant characteristics summary measures and HRQoL scores were computed as
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24 means and standard deviations for continuous (approximate) normally distributed
25
26 variables and frequencies and percentages for categorical variables. Normality of
27
28 distribution of continuous summary scales (all p -values >0.05) was assessed with the
29
30 Shapiro-Wilk test. Multiple linear regression analysis was performed to explore
31
32 associations between social support and HRQoL in patients in APS and to examine
33
34 whether perceived and ideal social support were associated with HRQoL. All analyses
35
36 were adjusted for age. All p -values were two-sided throughout and significance level
37
38 was set at 5% level. The data were analysed using SPSS version 21.
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45 Results

46 Participant characteristics

1 The majority of participants were from the United Kingdom (61.9%). Approximately a
2
3 quarter were from the United States (24.8%), and fewer from Australia (2.2%), Canada
4
5 (1.9%) and several other countries. Response rate was 60%. 274 patients completed
6
7 and returned the questionnaire survey out of a total of 443 individuals who were sent the
8
9 questionnaire survey link. Four questionnaires were discarded due to insufficient data
10
11 which resulted in 270 completed questionnaires being included in the analyses. Mean
12
13 patient age was 45.2±12.1 (range: 18-86 years). The majority of the patients were
14
15 female (84%; n=226) and 45% reported PAPS. Mean age for patients with PAPS was
16
17 42.6±11.6 years and for patients with SAPS 47.4±12.1 years. The mean time until
18
19 receiving a clinical diagnosis for APS was 48.5±87.3 months for PAPS and 75.8±106.4
20
21 months for SAPS patients. PAPS patients were prescribed a mean of 3±2.8 medications
22
23 while SAPS patients 7±5.2. SLE was reported by 43% (n=63) of SAPS participants. On
24
25 average, participants completed the survey five years post-diagnosis.
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32 Social support

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37 Frequency statistics were computed separately for actual and idea social support
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39 measures. The majority of patients indicated that they were perceived to be receiving
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41 emotional support such as listening (78.7%), understanding (59%), encouragement
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43 (55.6%) but not positive feedback (34% vs 66%) and willingness on behalf of their
44
45 family and friends to learn more about APS (44% vs 56%). In contrast, the majority of
46
47 patients perceived not to be receiving instrumental support such as help with childcare
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49 and housework/shopping (85.1% and 51.9% respectively), provision of transportation
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51 (69.8%), financial help (73.5%) and someone to accompany them to GP and hospital
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1 appointments (51.5%). Most of the information support, APS patients perceived to be
2
3 receiving was derived from the internet (82.1%) and support groups (65.7%) whereas a
4
5 minority of patients reported perceived informational support obtained from GPs
6
7 (26.9%) and TV or leaflets (14.2%) (Table 1).
8
9

10
11 The main social support (ideal) that patients would like to receive were: understanding
12
13 (67.9%), willingness to learn more about APS from family or friends (64.9%),
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15 information from GPs (74.3%), TV and leaflets (50%) (Table 1).
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19 HRQoL

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23 HRQoL scores were computed for all patients. Mean scores in six domains (RP, BP,
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25 GH, VT, SF and RE) were <60 which is the cut-off score reported to indicate highest
26
27 specificity for functional limitations [51]. In the remaining two SF-36 domains (PF and
28
29 MH), mean scores were >60. The mean HRQoL score was 64.4±20.6. Comparison
30
31 between PAPS and SAPS patients showed poorer HRQoL scores for the PAPS group (<
32
33 60) in two domains (GH and VT) and better HRQoL scores (>60) in the remaining six
34
35 domains (PF, RP, BP, RE, MH and SF) with a mean of 65.15±31.1. For SAPS patients,
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37 mean scores were <60 in seven of the SF-36 domains (RP, BP, GH, RE, VT, MH and
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39 SF) and >60 in one domain (PF) with a mean of 60.25±23.1.
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46 Relationship between social support and HRQoL in APS

47 Perceived social support and HRQoL

1 Perceived social support was assessed on three levels: emotional, instrumental and
2
3 informational. On the perceived emotional level, results showed that higher levels of
4
5 encouragement were related to better physical functioning (B=7.77, p<0.01; 95%CI:
6
7 2.25, 13.29), better role physical functioning (B=15.83; p<0.01; 95%CI: 3.96, 27.70)
8
9 and better general health (B=5.62; p<0.01; 95%CI: 1.02, 10.22) while less
10
11 understanding from friends and family were associated with lower levels of vitality
12
13 (B=-6.22, p<0.01; 95%CI:-10.91, -1.53). Lower provision of positive feedback was
14
15 associated with poorer role emotional functioning (B=-13.88, p<0.05; 95%CI: -26.51, -
16
17 1.25) and poorer mental health (B=-7.99, p<0.001; 95%CI: -12.52, -3.47) and similarly
18
19 lower degree of willingness to learn more about APS was also related to poorer mental
20
21 health (B=-4.27, p<0.05; 95%CI: -8.51, -0.02) (Table 2).
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29 Results on perceived instrumental support indicated that lower provision of help with
30
31 childcare was related to more limited social functioning (B=-9.21, p<0.05; 95%CI: -
32
33 18.49, 0.07) whereas more support in terms of helping with housework and/or shopping
34
35 were associated with better physical functioning (B=13.50, p<0.001; 95%CI: 8.17,
36
37 18.84), role physical (B=18.64, p<0.01; 95%CI: 6.88, 30.40), lower bodily pain
38
39 (B=17.52, p<0.001; 95%CI: 11.15, 23.90), better general health (B=10.40, p<0.001;
40
41 95%CI: 5.94, 14.87), higher levels of vitality (B=8.85, p<0.001; 95%CI: 4.30, 13.39),
42
43 and better social functioning (B=9.22, p<0.01; 95%CI: 2.71, 15.73). Provision of
44
45 transportation was associated with better HRQoL in all domains except for mental
46
47 health while financial help was associated with better physical functioning (B=7.93,
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49 p<0.01; 95%CI: 1.70, 14.16) and lower bodily pain (B=9.31, p<0.05; 95%CI: 1.77,
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1 16.85). Attendance at **General Practitioner (GP)** and hospital appointments was also
2
3 related to better HRQoL in the domains physical functioning (B=9.20, p<0.001; 95%CI:
4
5 3.71, 14.69), bodily pain (B=9.74, p<0.01; 95%CI: 3.09, 16.39), general health (B=5.53,
6
7 p<0.05; 95%CI: 0.92, 10.14), and role emotional (B=12.14, p<0.05; 95%CI: 0.12,
8
9 24.15) (see Table 3).
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13
14 Perceived informational support was associated with HRQoL in terms of information
15
16 provided by GPs, support groups, and consultants/charity. Less information provided by
17
18 GPs was associated with more limited physical functioning (B=-6.30, p<0.05; 95%CI: -
19
20 12.52, -0.08), and poorer role physical performance (B=-19.37, p<0.01; 95%CI: -32.63,
21
22 -6.11), more support provided by support groups was related to better social functioning
23
24 (B=8.06, p<0.05; 95%CI: 1.17, 14.94) and less information from consultants/charity
25
26 was related to poorer general health (B=-8.67, p<0.05; 95%CI: -16.72, -0.61) (Table 4).
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31 Ideal social support and HRQoL 32 33

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35 Ideal social support was also assessed on three levels: emotional, instrumental and
36
37 informational. Patients reported the levels of social support they felt they still wanted to
38
39 receive based on their needs. **People who felt they still needed someone to listen to their**
40
41 **concerns and worries would be more likely to have better physical functioning if they**
42
43 **had this support** (B=5.80, p<0.05; 95%CI: 0.26, 11.34) and higher levels of vitality
44
45 (B=6.91, p<0.01; 95%CI: 2.32, 4.51). The need for understanding was linked to better
46
47 HRQoL except for bodily pain and role emotional and the need for more frequent
48
49 encouragement was associated with better physical functioning (B=7.78, p<0.01;
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51 95%CI: 2.30, 13.26), role physical (B=14.46, p<0.05; 95%CI: 2.66, 26.26), and greater
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1 vitality (B=5.16, $p<0.05$; 95%CI: 0.56, 9.77). Greater provision of positive feedback
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3 was associated with better role physical (B=16.35, $p<0.01$; 95%CI: 4.59, 28.12), greater
4
5 vitality (B=5.29, $p<0.05$; 95%CI: 0.69, 9.90), and role emotional (B=13.13, $p<0.05$;
6
7 95%CI: 1.23, 25.04). Patients' greater need for their family and friends to learn more
8
9 about APS was related to better role physical (B=15.99, $p<0.01$; 95%CI: 3.62, 28.36),
10
11 and greater vitality (B=7.21, $p<0.01$; 95%CI: 2.42, 12.01) (Table 5).
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16 Ideal instrumental support was associated with better HRQoL in all domains in terms of
17
18 provision of help with housework and/or shopping and attendance at GP and hospital
19
20 appointments (see Table 6). The need for provision of transportation was related to
21
22 better HRQoL in all domains except role physical and role emotional (see Table 6).
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26
27 There was an association between greater informational support provided by and better
28
29 role emotional (B=19.05, $p<0.01$; 95%CI: 5.39, 32.70) and better mental health
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31 (B=6.29, $p<0.01$; 95%CI: 1.35, 11.23), while information from support groups was
32
33 associated with greater vitality (B=5.85, $p<0.01$; 95%CI: 1.23, 10.46) (Table 7).
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37
38 Discrepancy values between perceived and ideal social support
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41 In order to examine discrepancy between actual and ideal social support in APS
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43 patients, the total values of each were computed for all three types of social support, i.e.
44
45 emotional, instrumental and informational. Following that, a discrepancy value was
46
47 obtained by subtracting the total actual support values from the total ideal support
48
49 values for each of the three types of social support. The results indicated that the mean
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51 of the discrepancy value between actual and ideal emotional support ($M=.064$;
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1 $SD=2.42$; $N=268$) as well as the mean of the discrepancy between actual and ideal
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3 instrumental support ($M=-.011$; $SD=1.24$; $N=268$) were relatively small. In contrast, the
4
5 mean of the discrepancy between ideal and actual informational support ($M=.23$;
6
7 $SD=1.58$; $N=268$) was large. Thus, it would seem that the informational support APS
8
9 patients perceive to be receiving is far smaller than the support they ideally would need
10
11 to be receiving. As far as informational and instrumental support was concerned, the
12
13 ideal levels of social support did not appear to differ significantly from the perceived
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18 levels.

21 Discussion

24 This is the first study to examine associations between social support and HRQoL in
25
26 patients with APS. HRQoL in individuals living with rheumatic and autoimmune
27
28 diseases is affected to a significant extent [4, 5] with social support playing a significant
29
30 role [18, 37-39]. However, very little research has been conducted into HRQoL and
31
32 social support in patients with APS so far, to our knowledge. A variety of measures are
33
34 available to assess HRQoL including generic and disease-specific instruments. Generic
35
36 instruments can be employed across a range of different conditions while specific
37
38 instruments are disease-adapted. APS-related symptoms vary significantly and can have
39
40 an impact on patients' physical, social and emotional status [1].

47 Associations between perceived social support and HRQoL indicated that higher
48
49 provision of instrumental support was related to better HRQoL in terms of patients'
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51 physical and mental status. Higher perceived emotional support, on the other hand, led
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53 to better mental health status probably because increased support in this domain may
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1 contribute to patients getting more time to rest, socialize and experience less stress [43-
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3 46]. More perceived information provided by GPs was also related to better physical
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5 functioning and role physical, while information received from support groups was
6
7 related to better social functioning and less information from consultants and the charity
8
9 were related to poorer general health. This is in accordance with previous literature
10
11 which suggests that doctor-patient communication plays a significant role in patients'
12
13 health outcomes, quality of life, absence from work and treatment adherence [47]. It
14
15 could be assumed that because of insufficient information provided, patients are not
16
17 aware of the degree they should engage in various activities and this subsequently may
18
19 have a negative effect on their physical health due to increased or decreased
20
21 involvement.
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28 Similarly, higher ideal social support was also linked to better HRQoL. People who felt
29
30 they needed someone to listen to their concerns and worries were predicted to be more
31
32 likely to have better physical functioning such as fewer limitations in performing
33
34 physical activities, including bathing or dressing and experience greater vitality i.e.
35
36 feeling more energetic. This could be attributed to the fact that people might be likely
37
38 to receive some reassurance from their social circle that they are able to perform certain
39
40 activities following the expression of their concerns or offer them additional support
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42 which might contribute to decrease the burden that affect their energy levels. This was
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44 also reflect in the fact that the need for more frequent encouragement was associated
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46 with better physical while greater provision of positive feedback was associated with
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48 better role physical greater vitality, and role emotional.
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1 The need to feel understood by family and friends was linked to better HRQoL in all
2 domains except for physical pain and role emotional. Indeed, negative social responses,
3 particularly discounting (rejecting) and lack of understanding (not being
4 acknowledged), were associated with poorer health among patients with fibromyalgia
5 and rheumatoid arthritis [52]. Greater understanding would be derived from better
6 knowledge of APS by the patient's family and social circle. Results showed that
7 patients' greater need for their family and friends to learn more about APS was related
8 to better role physical, and greater vitality. Ideal instrumental support was associated
9 with better HRQoL in all domains in terms of provision of help with housework and/or
10 shopping and attendance at GP and hospital appointments and greater informational
11 support was associated with better role emotional and better mental health, while
12 information from support groups was associated with greater vitality.

13 In order to assess whether the social support that patients reported to be receiving was
14 significantly different to the support they felt they still needed, results did not show a
15 statistically significant difference for emotional and instrumental support. However,
16 there was a statistically significant difference between the informational support patients
17 reported to be receiving and the informational support they felt they were still lacking
18 highlighting the lack of awareness as well as health professional education and public
19 awareness about APS. More knowledge of APS provided by health care professionals
20 and the media might enhance patients' coping with the disease due to increased
21 education on aspects such as self-management, and medication and more effective and
22 helpful support from their families and friends.

1 There are some limitations to this study. First, the participants were members of the
2
3 **HSF** which might have increased the likelihood of receiving higher social support
4
5 compared to patients who do not belong to a charity. The data was based on self-report
6
7 thus environmental or emotional influences could not be controlled. Diagnosis of APS
8
9 could not be confirmed due to lack of access to patients' medical records or physical
10
11 and laboratory examination. The survey was cross-sectional which prevents detection
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13 of change over time and assessment of causal relationships. Findings cannot be
14
15 generalized due to the fact that the majority of patients were female and British.
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Ethnicity and culture has been suggested to affect perceived quality of life of individuals on dialysis after renal transplant with Asian renal patients perceiving HRQoL more negatively than white Europeans [47]. Factors such as major life events, for example death, divorce or severity of disease and depression status were not controlled for, any of which could potentially be related to poorer **HRQoL** in either group. Many factors such as bereavement, financial difficulties, depression and anxiety were also found to place a burden on **HRQoL** [11].

Strengths of this study, on the other hand, include a high response rate (60%), a satisfactory sample size providing sufficient power for analyses. The fact that a relatively non-researched patient population was assessed was a further advantage. Examination of all aspects of patients' well-being relating to the psychological, social and physical impact of APS and its influence by social support measures provided a more holistic approach and increased understanding of the degree and nature of the disease impact on patients' HRQoL.

1 Findings indicated that certain types of social support exert a significant influence on a
2
3 variety of domains in APS patients' HRQoL. Patients reported receiving insufficient
4
5 social support. By extension, this might be suggestive of the beneficial effect of social
6
7 support on HRQoL in patients with APS either through medication adherence or also
8
9 through more effective coping skills. Lack of support in terms of providing disease –
10
11 and medication-specific information has been associated with decreased medication
12
13 adherence in patients with autoimmune diseases [48]. In addition, increased
14
15 informational support especially by knowledgeable health professionals might improve
16
17 provision of support by patients' family and friends through reducing “invalidation”
18
19 [29]. Particularly, due to the multi-faceted nature of APS, as is true of most autoimmune
20
21 diseases, involving pain, disability, uncertainty about its progression and fear of
22
23 treatment effects and based on the current findings it can be suggested that a
24
25 combination of approaches and interventions could prove to be of great importance and
26
27 help in improving adjustment and coping with APS. This combination would need to
28
29 incorporate social approach and support from practitioners, family, friends, and co-
30
31 workers, as well as elements from both the bio-psychosocial and biomedical
32
33 frameworks [49, 50] tailored to the needs of APS patients. **Specific strategies could be**
34
35 **implemented through both primary and secondary care and include patient- and**
36
37 **family/friends-education sessions delivered by specialist nurses. These sessions could**
38
39 **provide disease- and treatment-specific information and self-management strategies**
40
41 **such as International Normalised Ratio (INR) measuring, dietary advice and pacing to**
42
43 **patients as well as disease-related information and ways of supporting their loved one in**
44
45 **coping more effectively with APS to families/friends of patients with APS.**
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1 **Acknowledgements**
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4 This research was supported by the Institute of Work, Health & Organisations of the
5
6 University of Nottingham. We are grateful to Ms. Kate Hindle, manager of the Hughes
7
8 Syndrome Foundation, for her invaluable help and support in conducting the study as
9
10 well as to the Hughes Syndrome London support group for their comments and helpful
11
12 suggestions. We would also like to acknowledge the significant contribution of all the
13
14 Hughes Syndrome Foundation members who participated in the present study. Last but
15
16 not least, we would like to express our gratitude to Professor Graham Hughes for his
17
18 encouragement, support and passion which inspired this research and motivated us
19
20 throughout the whole project. The views expressed here are the authors' and do not
21
22 necessarily reflect those of any other person or organization.
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32 **Funding**
33
34

35 This research ideal no specific grant from any funding agency in the public,
36
37 commercial, or not-for-profit sectors.
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45 **Conflict of interest** None.
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Table 1 Computed values of results for ideal and actual social support

| Actual Social Support - Emotional (n=268) | | | Ideal Social Support - Emotional (n=268) | | |
|--|-------------|-------------|---|-------------|-------------|
| | Yes | No | | Yes | No |
| Listening | 211 (78.7%) | 57 (21.3%) | Listening | 136 (50.7%) | 132 (49.3%) |
| Understanding | 158 (59%) | 110 (41%) | Understanding | 182 (67.9%) | 86 (32.1%) |
| Encouragement | 149 (55.6%) | 119 (44.4%) | Encouragement | 127 (47.4%) | 141 (52.6%) |
| Positive Feedback | 91 (34%) | 177 (66%) | Positive Feedback | 125 (46.6%) | 143 (53.4%) |
| Willingness to learn more about APS | 118 (44%) | 150 (56%) | Willingness to learn more about APS | 174 (64.9%) | 94 (35.1%) |
| Actual Social Support - Instrumental (n=268) | | | Ideal Social Support - Instrumental (n=268) | | |
| Help with childcare | 40 (14.9%) | 228 (85.1%) | Help with childcare | 41 (15.3%) | 227 (84.7%) |
| Help with housework/ shopping | 129 (48.1%) | 139 (51.9%) | Help with housework/ shopping | 121 (45.1%) | 147 (54.9%) |
| Provision of transportation | 81 (30.2%) | 187 (69.8%) | Provision of transportation | 66 (24.6%) | 202 (75.4%) |
| Financial help | 71 (26.5%) | 197 (73.5%) | Financial help | 68 (25.4%) | 200 (74.6%) |
| Attendance GPs/hospital appt | 131 (48.9%) | 137 (51.5%) | Attendance GPs/hospital appt | 109 (40.7%) | 159 (59.3%) |
| Actual Social Support - Informational (n=268) | | | Ideal Social Support - Informational (n=268) | | |
| Information provided by GPs | 72 (26.9%) | 196 (73.1%) | Information provided by GPs | 199 (74.3%) | 69 (25.7%) |
| Information provided on the internet | 220 (82.1%) | 48 (17.9%) | Information provided on the internet | 119 (44.4%) | 149 (55.6%) |
| Information provided by support groups | 176 (65.7%) | 92 (34.3%) | Information provided by support groups | 119 (44.4%) | 149 (55.6%) |
| Information provided on TV/ leaflets | 38 (14.2%) | 229 (85.4%) | Information provided on TV/ leaflets | 134 (50.0%) | 134 (50.0%) |
| Information provided by consultants/charity (n=20) | 19 (7.1%) | 1 (0.4%) | Information provided by consultants/charity (n=42) | 41 (15.2%) | 1 (0.4%) |

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Table 2 Associations between perceived emotional support and HRQoL in APS

| <i>Perceived Social Support – Emotional (n= 270)</i> | | | | | |
|--|-------------------------|----------------------------|--------------------------|-----------------------------|--|
| <i>SF-36 domains</i> | <i>Listening</i> | <i>Understanding</i> | <i>Encouragement</i> | <i>Positive Feedback</i> | <i>Willingness to learn more about APS</i> |
| | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| PC | | | | | |
| Physical functioning | -0.50 (-7.29, 6.30) | -2.62 (-8.31, 3.06) | 7.77** (2.25, 13.29) | 0.43 (-5.47, 6.33) | 4.21 (-1.21, 9.62) |
| Role physical | 5.90 (-8.68, 20.48) | -5.60 (-17.79, 6.60) | 15.83** (3.96, 27.70) | 4.84 (-7.82, 17.50) | 0.32 (-11.37, 12.01) |
| Bodily pain | 2.97 (-5.25, 11.19) | -2.23 (-9.08, 4.62) | 6.62 (-0.08, 13.33) | 0.43 (-6.68, 7.53) | 1.35 (-5.21, 7.91) |
| General health | -2.74 (-8.40, 2.92) | -1.74 (-6.46, 2.98) | 5.62** (1.02, 10.22) | -0.45 (-5.35, 4.46) | 0.57 (-3.96, 5.10) |
| MC | | | | | |
| Vitality | -3.66 (-9.35, 2.03) | -6.22** (-10.91, -1.53) | 1.74 (-2.94, 6.41) | -4.23 (-9.13, 0.68) | -3.62 (-8.15, 0.92) |
| Social functioning | -3.82 (-11.88, 4.25) | -4.60 (-11.30, 2.11) | 1.82 (-4.80, 8.43) | -3.64 (-10.61, 3.33) | -3.03 (-9.47, 3.41) |
| Role emotional | -9.60 (-24.24, 5.03) | -7.56 (-19.83, 4.71) | -2.45 (-14.55, 9.65) | -13.88* (-26.51, -1.25) | -10.38 (-22.06, 1.31) |
| Mental health | -3.44 (-8.78, 1.90) | -3.99 (-8.43, 0.44) | -2.57 (-6.95, 1.81) | -7.99*** (-12.52, -3.47) | -4.27* (-8.51, -0.02) |

Multiple Linear Analysis examining the association between perceived emotional support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 3 Associations between perceived instrumental support and HRQoL in APS

| <i>Perceived Social Support – Instrumental (n= 270)</i> | | | | | |
|---|----------------------------|--------------------------------------|------------------------------------|-------------------------|---|
| <i>SF-36 domains</i> | <i>Help with childcare</i> | <i>Help with housework/ shopping</i> | <i>Provision of transportation</i> | <i>Financial help</i> | <i>Attendance at GPs/ hospital appointments</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | 0.24 (-7.63, 8.10) | 13.50*** (8.17, 18.84) | 15.03*** (9.26, 20.81) | 7.93** (1.70, 14.16) | 9.20*** (3.71, 14.69) |
| Role physical | 1.09 (-15.83, 17.99) | 18.64** (6.88, 30.40) | 20.20** (7.40, 32.99) | 11.21 (-2.28, 26.69) | 8.92 (-3.06, 20.91) |
| Bodily pain | 0.15 (-9.50, 9.81) | 17.52*** (11.15, 23.90) | 14.59*** (7.51, 21.67) | 9.31* (1.77, 16.85) | 9.74** (3.09, 16.39) |
| General health | 0.75 (-5.82, 7.31) | 10.40*** (5.94, 14.87) | 6.03* (1.03, 11.03) | 3.88 (-1.36, 9.12) | 5.53* (0.92, 10.14) |
| <i>MC</i> | | | | | |
| Vitality | -0.08 (-6.69, 6.52) | 8.85*** (4.30, 13.39) | 8.48*** (3.50, 13.46) | 1.67 (-3.62, 6.95) | 1.18 (-3.50, 5.87) |
| Social functioning | -9.21* (-18.49, 0.07) | 9.22** (2.71, 15.73) | 10.43** (3.34, 17.52) | 2.10 (-5.39, 9.58) | 5.26 (-1.34, 11.86) |
| Role emotional | -8.29 (-25.25, 8.67) | 5.59 (-6.45, 17.62) | 14.95* (1.97, 27.92) | 7.15 (-6.44, 20.74) | 12.14* (0.12, 24.15) |
| Mental health | -3.26 (-9.44, 2.93) | 2.46 (-1.91, 6.83) | 3.63 (-1.12, 8.38) | 0.33 (-4.63, 5.30) | -0.22 (-4.62, 4.18) |

Multiple Linear Analysis examining the association between perceived instrumental support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 4 Associations between perceived informational support and HRQoL in APS

| <i>Perceived Social Support – Informational (n= 270)</i> | | | | | |
|--|------------------------------------|---|---|--|--|
| <i>SF-36 domains</i> | <i>Information provided by GPs</i> | <i>Information provided on the internet</i> | <i>Information provided by support groups</i> | <i>Information provided on TV/leaflets</i> | <i>Information provided by consultants/charity</i> |
| | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| PC | | | | | |
| Physical functioning | -6.30* (-12.52, -0.08) | -2.17 (-9.50, 5.16) | 4.20 (-1.65, 10.05) | 4.42 (-3.49, 12.33) | -3.02 (-12.74, 6.69) |
| Role physical | -19.37** (-32.63, -6.11) | -3.52 (-19.16, 12.11) | 6.93 (-5.65, 19.51) | 8.03 (-8.99, 25.05) | -14.63 (-35.45, 6.20) |
| Bodily pain | -6.12 (-13.63, 1.39) | 3.02 (-5.65, 11.69) | 4.57 (-2.50, 11.64) | 6.15 (-3.37, 15.66) | -5.52 (-17.19, 6.15) |
| General health | -0.50 (-5.71, 4.70) | 0.41 (-5.61, 6.43) | 2.07 (-2.80, 6.95) | 2.89 (-3.66, 9.44) | -8.67* (-16.72, -0.61) |
| MC | | | | | |
| Vitality | -4.59 (-9.80, 0.62) | 1.50 (-4.56, 7.56) | 0.65 (-4.26, 5.56) | -2.40 (-8.99, 4.19) | -1.18 (-9.35, 6.99) |
| Social functioning | -6.79 (-14.16, 0.58) | -2.63 (-11.20, 5.94) | 8.06* (1.17, 14.94) | 8.86 (-0.42, 18.14) | -9.34 (-20.85, 2.18) |
| Role emotional | -11.02 (-24.49, 2.40) | -13.45 (-29.21, 2.31) | 5.10 (-7.57, 17.76) | -2.99 (-20.12, 14.13) | -8.62 (-29.59, 12.36) |
| Mental health | -1.12 (-6.03, 3.80) | -4.08 (-9.74, 1.59) | -1.09 (-5.69, 3.52) | -1.71 (-7.90, 4.48) | -3.72 (-11.38, 3.94) |

Multiple Linear Analysis examining the association between perceived informational support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 5 Associations between ideal emotional support and HRQoL in APS

| <i>Ideal Social Support – Emotional (n= 270)</i> | | | | | |
|--|------------------------|--------------------------|-------------------------|--------------------------|--|
| <i>SF-36 domains</i> | <i>Listening</i> | <i>Understanding</i> | <i>Encouragement</i> | <i>Positive Feedback</i> | <i>Willingness to learn more about APS</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | 5.80* (0.26, 11.34) | 6.21* (0.31, 12.10) | 7.78** (2.30, 13.26) | 3.99 (-1.54, 9.54) | 4.91 (-0.89, 10.71) |
| Role physical | 5.76 (-6.21, 17.73) | 14.27* (1.61, 26.93) | 14.46* (2.66, 26.26) | 16.35** (4.59, 28.12) | 15.99** (3.62, 28.36) |
| Bodily pain | 0.23 (-6.50, 6.96) | 5.87 (-1.26, 12.99) | 2.66 (-4.04, 9.35) | -0.005 (-6.70, 6.69) | 0.75 (-6.27, 7.76) |
| General health | 2.06 (-2.57, 6.70) | 5.66* (0.77, 10.56) | 3.64 (-0.96, 8.24) | 1.41 (-3.21, 6.02) | 2.30 (-2.53, 7.14) |
| <i>MC</i> | | | | | |
| Vitality | 6.91** (2.32, 4.51) | 9.48*** (4.64, 14.32) | 5.16* (0.56, 9.77) | 5.29* (0.69, 9.90) | 7.21** (2.42, 12.01) |
| Social functioning | 5.55 (-1.03, 12.12) | 7.53* (0.55, 14.51) | 6.31 (-0.23, 12.85) | 4.10 (-2.47, 10.66) | 4.77 (-2.10, 11.65) |
| Role emotional | 9.91 (-2.10, 21.92) | 4.88 (-7.95, 17.72) | 7.69 (-4.29, 19.67) | 13.13* (1.23, 25.04) | 8.41 (-4.15, 20.96) |
| Mental health | 2.32 (-2.06, 6.69) | 6.17** (1.57, 10.78) | 2.55 (-1.81, 6.90) | 1.68 (-2.68, 6.04) | 1.74 (-2.83, 6.30) |

Multiple Linear Analysis examining the association between ideal emotional support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 6 Associations between ideal instrumental support and HRQoL in APS

| <i>Ideal Social Support – Instrumental (n= 270)</i> | | | | | |
|---|----------------------------|--------------------------------------|------------------------------------|-------------------------|---|
| <i>SF-36 domains</i> | <i>Help with childcare</i> | <i>Help with housework/ shopping</i> | <i>Provision of transportation</i> | <i>Financial help</i> | <i>Attendance at GPs/ hospital appointments</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | -0.64 (-8.56, 7.28) | 10.77*** (5.33, 16.20) | 11.28*** (4.99, 17.58) | 3.67 (-2.72, 10.07) | 11.81*** (6.29, 17.33) |
| Role physical | -1.50 (-18.54, 15.54) | 20.03*** (8.29, 31.77) | 12.93 (-0.83, 26.69) | 11.59 (-2.05, 25.24) | 18.78** (6.77, 30.79) |
| Bodily pain | -4.52 (-14.13, 5.09) | 12.01*** (5.45, 18.58) | 10.63** (2.95, 18.31) | 4.50 (-3.16, 12.17) | 9.87** (3.12, 16.62) |
| General health | 0.75 (-5.82, 7.31) | 8.48*** (3.95, 13.00) | 5.58* (0.25, 10.91) | 2.41 (-2.91, 7.73) | 7.36** (2.71, 12.01) |
| <i>MC</i> | | | | | |
| Vitality | 3.91 (-2.68, 10.50) | 11.69*** (7.24, 16.14) | 9.35*** (4.07, 14.64) | 5.17 (-0.15, 10.49) | 9.86*** (5.25, 14.47) |
| Social functioning | 0.83 (-8.52, 10.18) | 11.82*** (5.37, 18.28) | 11.88*** (4.37, 19.40) | 1.64 (-5.94, 9.22) | 9.94** (3.31, 16.58) |
| Role emotional | 0.97 (-16.15, 18.08) | 16.75** (4.85, 28.66) | 12.12 (-1.72, 25.96) | 10.53 (-3.26, 24.32) | 14.28* (2.07, 26.48) |
| Mental health | 0.83 (-5.37, 7.03) | 4.73* (0.38, 9.07) | 5.79* (0.77, 10.82) | 3.56 (-1.45, 8.57) | 4.46* (0.02, 8.90) |

Multiple Linear Analysis examining the association between ideal instrumental support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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Table 7 Associations between ideal informational support and HRQoL in APS

| <i>Ideal Social Support – Informational (n= 270)</i> | | | | | |
|--|------------------------------------|---|---|--|--|
| <i>SF-36 domains</i> | <i>Information provided by GPs</i> | <i>Information provided on the internet</i> | <i>Information provided by support groups</i> | <i>Information provided on TV/leaflets</i> | <i>Information provided by consultants/charity</i> |
| PC | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) | B (95% CI) |
| Physical functioning | 2.23 (-4.18, 8.64) | -0.64 (-6.27, 4.99) | 4.18 (-1.39, 9.75) | 3.20 (-2.37, 8.76) | -4.12 (-8.82, 0.59) |
| Role physical | -0.63 (-14.40, 13.15) | -1.76 (-13.84, 10.32) | 6.04 (-5.94, 18.01) | 11.41 (-0.48, 23.30) | 1.37 (-8.80, 11.53) |
| Bodily pain | 1.99 (-5.68, 9.66) | -1.46 (-8.25, 5.32) | 2.19 (-4.55, 8.92) | 5.00 (-1.69, 11.69) | -0.77 (-6.62, 5.07) |
| General health | 0.42 (-4.87, 5.71) | 1.75 (-2.91, 6.42) | 2.68 (-1.95, 7.31) | 1.01 (-3.63, 5.64) | -0.79 (-4.74, 3.16) |
| <i>MC</i> | | | | | |
| Vitality | 3.34 (-1.97, 8.65) | 4.24 (-0.44, 8.91) | 5.85** (1.23, 10.46) | 2.40 (-2.25, 7.06) | -1.33 (-5.30, 2.65) |
| Social functioning | 2.29 (-5.24, 9.82) | 0.20 (-6.46, 6.85) | 3.18 (-3.43, 9.78) | 1.69 (-4.91, 8.29) | -1.93 (-7.55, 3.70) |
| Role emotional | 19.05** (5.39, 32.70) | -2.36 (-14.52, 9.81) | 7.70 (-4.34, 19.75) | 11.27 (-0.70, 23.25) | -2.34 (-12.55, 7.87) |
| Mental health | 6.29** (1.35, 11.23) | 3.49 (-0.91, 7.88) | 3.81 (-0.55, 8.17) | 2.19 (-2.18, 6.55) | -1.01 (-4.74, 2.72) |

Multiple Linear Analysis examining the association between ideal informational support and HRQoL variables adjusted for age; SF-36: Medical Outcomes Study Short-Form 36; PC: physical component; MC: mental component; CI: confidence intervals * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$

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