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Predictors of the likelihood that patients with rheumatoid arthritis will communicate information about rheumatoid arthritis risk to relatives: A quantitative assessment

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ABSTRACT

First-degree relatives (FDRs) of people with rheumatoid arthritis (RA) are increasingly recruited to prediction and prevention studies. Access to FDRs is usually via their proband with RA. Quantitative data on predictors of family risk communication are lacking. RA patients completed a questionnaire assessing likelihood of communicating RA risk information to their FDRs, demographic variables, disease impact, illness perceptions, autonomy preferences, interest in FDRs taking a predictive test for RA, dispositional openness, family functioning, and attitudes towards predictive testing. Ordinal regression examined associations between patients' characteristics and their median likelihood of communicating RA risk to FDRs. Questionnaires were completed by 482 patients. The majority (75.1%) were likely/extremely likely to communicate RA risk information to FDRs, especially their children. Decision-making preferences, interest in FDRs taking a predictive test, and beliefs that risk knowledge would increase people's empowerment over their health increased patients' odds of being likely to communicate RA risk information to FDRs. Beliefs that risk information would cause stress to their relatives decreased odds that patients would be likely to communicate RA risk. These findings will inform the development of resources to support family communication about RA risk.

1. Introduction

Rheumatoid arthritis (RA) is a chronic condition affecting ≈ 1% of the population.[1] Early treatment improves outcomes.[1,2] There is increasing research focus on those at risk of developing RA, to facilitate the development of preventive interventions.[3–5].

First-degree relatives (FDRs) of RA patients have an increased risk of developing RA by approximately 3–5 fold.[6] Environmental risk factors

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may further increase risk of RA for FDRs.[7–9] Several prospective observational studies are recruiting FDRs to develop predictive algorithms for RA.[10–12].

Integral to the success of predictive and preventive strategies for FDRs is the family member with RA, as access to FDRs is usually obtained via that proband. If patients are unwilling or unable to pass on information to their FDRs about their risk of developing RA, or about opportunities for predictive testing or preventive strategies, then access to this group may be restricted. It is therefore important to understand the process and determinants of family communication about RA risk to develop effective strategies to support effective communication and access to FDRs.

Previous studies examining risk communication in families across other disease areas including cancer and cardiovascular disease (CVD) [13–16] identified factors influencing patients' likelihood of communicating disease risk to relatives. These included closeness with their relatives, perceived responsibility to provide this information, and perceived receptiveness of relatives to this information. Females were more likely than males to communicate and receive risk information. [13] Patients who were unlikely to communicate risk information highlighted a desire to protect their relatives from anxiety-provoking information.[13,14] Whilst these findings provide insight into the process of family communication about risk, the diseases that have been studied to date are often perceived as more severe than RA.[17] There is also an increased public awareness of these diseases compared to RA. [18,19] It is therefore important to understand the process and determinants of family risk communication for those at risk of RA.

One qualitative study investigated RA patients' perspectives on communicating RA risk information to their FDRs.[20] Patients expressed willingness to communicate with FDRs about their risk of RA. However, they described a process of selecting which relatives to communicate with. This was based on perceived receptivity of their FDRs, FDRs' likelihood to act on this information, and patients' feelings of guilt and responsibility around passing on a hereditary predisposition to their FDRs. Reasons patients provided for not wanting to communicate risk information included a lack of closeness with their FDRs and wanting to protect their FDRs from unnecessary anxiety. No quantitative studies to date have examined determinants of family communication about RA risk, therefore the aim of this study is to assess predictors of RA patients' reported likelihood of communicating RA risk information to their FDRs.

2. Method

2.1. Design

A cross-sectional survey was provided to patients diagnosed with RA which assessed their likelihood of communicating RA risk to each of their FDRs (primary outcome), and potential demographic and psychosocial predictors of their likelihood to communicate risk information.

2.2. Procedure

Patients with a confirmed diagnosis of RA were identified via rheumatology outpatient clinics in the West Midlands, England and were recruited between March 2017 and January 2020. Patients were eligible if they (i) had received a diagnosis of RA (satisfying the 2010 American College of Rheumatology/ European League Against Rheumatism classification criteria [21]) at least six months before they were approached to take part in the study; (ii) were aged 18 years or over; (iii) had one or more FDRs (biological offspring or full siblings); and (iv) could complete the printed survey in English. All patients provided written informed consent.

Patients were introduced to the study by a member of their healthcare team during a scheduled rheumatology outpatient clinic visit and were provided with a survey, and a freepost envelope to return the completed anonymous survey to the research team. Patients were advised they could take the survey pack home and decide whether to participate in their own time.

The study was approved by the Research Ethics Committee (Berkshire B): 16/SC/0369.

2.3. Measures

2.3.1. Primary outcome measure

Patients were asked to identify their relationship to each FDR (daughter, son, sister or brother), and their likelihood of communicating RA risk information to each of those FDRs ("How likely would you be to pass on information to this relative about their risk of developing rheumatoid arthritis?" assessed on a 5-point Likert scale from 0 (extremely unlikely) to 4 (extremely likely), with higher scores indicating increased likelihood).

2.3.2. Measures of potential predictors of patients' likelihood to communicate RA risk information to their FDRs

Selection of measures was informed by a literature review of research on family communication of disease risk, which identified: demographic factors [22,23]; disease impact [24,25]; illness perceptions [26,27]; health literacy and numeracy [28,29]; preferences for autonomy in health-related information seeking and decision-making [14,30]; coping styles [26,31]; dispositional optimism [32]; dispositional openness [33]; family functioning [27,34]; interest in relatives taking a predictive test [31,35] and; attitudes towards predictive testing [13,36] as potential predictor variables. Brief versions of relevant measures were included where available in response to patient partner assessment of cognitive burden for participants. Patients reported their gender, age, ethnicity, postcode (used to calculate the Index of Multiple Deprivation score, where a score of 1 indicates the most deprived areas and a score of 10 indicates the least deprived areas), employment status, highest level of education, smoking status, years with RA and current treatment for RA.

The following measures were also completed:

(1) Rheumatoid Arthritis Impact of Disease (RAID) scale; assessing RA impact over the last week across seven domains: pain, ability, fatigue, sleep, physical wellbeing, emotional wellbeing and coping). Each domain was measured on an 11-point scale from 0 (no impact) to 10 (extreme impact). Higher scores indicate worse disease status.[37]
(2) The Brief Illness Perceptions Questionnaire (Brief IPQ); measuring patients' RA related illness perceptions in eight domains: consequences, timeline, personal control, treatment control, identity, concern, understanding and emotion. Items were scored on an 11-point scale, with higher scores indicating a more threatening view of RA.[38,39]
(3) The single item literacy screener (SILS); assessing patients' health literacy. Responses were scored on a 5-point Likert scale from 0 (never) to 4 (always). Scores above 2 indicate some difficulty reading health-related material.[40]
(4) The three-item subjective numeracy scale (SNS-3); measuring patients' self-reported ability to understand numerical information. Each item was scored on a 6-point Likert scale, with higher scores indicating stronger perceived mathematical ability. [41].
(5) The Autonomy Preference Index (API); measuring health-related decision-making (six items) and information-seeking preferences (eight items).[42] Each item was measured on a 5-point Likert scale from 0 (strongly disagree) to 4 (strongly agree). For each subscale, scores were converted into a scale from 0 to 100, with higher scores indicate greater autonomy preferences. [42]
(6) The Brief Approach/Avoidance Coping Questionnaire; measuring approach/avoidant coping style in stressful situations in three domains: cognitive, socioemotional and action-related. [43]

Items were measured using a 5-point Likert scale from 0 (strongly disagree) to 4 (strongly agree). Total scores range from 0 to 48, with higher scores indicating higher approach or lower avoidance coping styles. [43]

(7) Dispositional optimism, assessed using the three items from the Life Orientation Test-Revised (LOT-R). These items were measured using a 5-point Likert scale from 0 (strongly disagree) to 4 (strongly agree). Higher scores indicate increased optimism. [44]

(8) Dispositional openness measured patients' general disclosure of information using a 5-point Likert scale from 0 (strongly disagree) to 4 (strongly agree). Higher scores indicate increased openness. [45]

(9) The General Functioning Subscale of the McMaster Family Assessment Device; measuring family functioning across six domains: general problem solving, communication, roles, affective responses, affective involvement and behavioural control. [46] These items were measured on a 4-point Likert scale ranging from 0 (strongly disagree) to 3 (strongly agree). Scores above 2 indicate good family functioning.

(10) Rating scale of patients' interest in their children and/or siblings taking a predictive test within 6 months was assessed on a 4-point Likert scale from 0 (no definitely not) to 3 (yes definitely). Higher scores indicate increased interest.

(11) Twenty-three attitudinal statements measuring perceived advantages (12 items) and disadvantages (11 items) of "someone finding out how likely they are to develop rheumatoid arthritis in the future". Sixteen of these items (seven advantages and nine disadvantages) were adapted from Cameron et al. [47], with an additional seven items (five advantages and two disadvantages) based on themes identified in previous qualitative investigations. [18,48–50] These items are assessed on a 5-point Likert scale from 0 (strongly disagree) to 4 (strongly agree).

(12) Thirty-two statements measuring possible reasons why a patient may be likely / unlikely to pass on RA risk information to their relatives. These statements were informed by previous qualitative findings.[17] Items were assessed on a 5-point Likert scale from 0 (definitely does not apply) to 4 (definitely applies).

2.4. Analysis

Statistical analyses were performed using IBM SPSS Statistics version 27.0.

Descriptive statistics were used to summarise demographic and psychosocial characteristics, variance in patients' likelihood of communicating risk information to each of their FDRs, and reasons patients were likely/ unlikely to communicate RA risk information to their relatives. Principal components analysis (PCA) with direct oblimin rotation was conducted to reduce the 23 attitudinal items into a smaller number of underlying factors. Original scores for each item were multiplied by factor loadings to obtain a weighted score. From this, a mean score was calculated.

Within-person variance on the risk communication likelihood measure was examined using the Friedman test to detect differences in scores across FDR responses.

Kruskal-Wallis H and Mann-Whitney U tests were performed to assess the effects of categorical variables on patients' reported likelihood of communicating RA risk to their FDRs. Spearman's rank correlations were used to investigate associations between ordinal predictor variables and likelihood of communicating RA risk. Wilcoxon signed-rank tests were conducted on patients who reported having both children and siblings, as well as patients who reported having both male and female relatives, to examine differences in patients' likelihood of communicating RA risk information to their children compared to their siblings, and to male relatives compared to female relatives. All predictor variables with a significance level < 0.1 informed an ordinal logistic regression model using backward elimination, with likelihood of communicating RA risk recoded as 'extremely likely to communicate RA risk information', 'likely to communicate RA risk information', or 'unlikely to communicate RA risk information'(scored from 0 to 2, respectively). The dependent variable was recoded in this manner due to the small number of responses occurring in the 'extremely unlikely' ($n = 26$), 'unlikely' ($n = 40$) and 'neither likely nor unlikely' ($n = 54$) groups. These three groups were treated as 'unlikely to communicate RA risk information'. Multicollinearity among the predictor variables used in the ordinal regression was assessed using variance inflation factors (VIFs) and a correlation matrix.

For patients' reported likelihood of communicating RA risk information to their FDRs, the median score across all FDRs was calculated for each patient and used as the primary outcome in Kruskal-Wallis H, Mann-Whitney U, Spearman's rank and ordinal regression analyses. For Wilcoxon signed-rank tests, the median scores across each FDR group (male, female, child, sibling) were calculated for each patient and compared.

2.5. Sample size calculation

A sample size of 480 patients would provide 95% confidence that an estimate of the proportion of positive and negative responses for the primary outcome variable was within 0.046 of the true value.

2.6. Patient and public involvement

Three patient research partners (PRPs) contributed to survey development, commenting on drafts of the protocol, study documents and surveys (via email), and attending a focus group to discuss survey design and content. A detailed description of the involvement of these patient research partners and their impact on the study has been reported in a previous paper.[51].

3. Results

Surveys were provided to 1720 patients. 482 of these patients returned a survey. The median age for this sample was 65 years, 72% of participants were female and 50% were retired. Patients reported having had a diagnosis of RA for a median of 10 years, and most reported taking conventional synthetic DMARDs and/or glucocorticoids to manage their condition (89%) (Table 1).

3.1. Patients' likelihood of communicating RA risk information to their FDRs

Most patients reported being "likely" or "extremely likely" to communicate RA risk information to their FDRs (38.2% and 36.9%, respectively) (Table 2). 81.2% of patients reported being "likely" or "extremely likely" to communicate RA risk information to their children, 69.3% to their siblings, 75.8% to male relatives, and 77.2% to female relatives.

When examining within-person variance of patients' likelihood to communicate RA risk information, the range between patients' highest and lowest scores across their FDRs was low for most patients. Where patients' likelihood to communicate RA risk is scored from 0 (extremely unlikely) to 4 (extremely likely), the range between patients' scores was 0 for 72% of patients, 1 for 11.3%, 2 for 6.8%, 3 for 5.3% and 4 for 4.7%. The Friedman test for within-person variance was significant ($p < 0.001$), indicating that there are significant differences in the distribution of scores across patients' responses for each FDR.

The 190 patients who reported their likelihood of communicating RA risk to both children and siblings were more likely to communicate about risk to their children compared to their siblings ($p < 0.001$) (Table 3). Of the 221 patients who reported their likelihood of communicating RA risk information to both male and female relatives,

Table 1
Descriptive statistics and univariate analyses for patients' characteristics and associations with their median likelihood of communicating RA risk to FDRs ($n = 482$).

Patient characteristics	Descriptive statistics				
	Frequency for patients' characteristics	Medians (IQRs) for patients' characteristics	Medians (IQRs) for patients' likelihood of communicating risk	Test statistics	P value
Age (years) (N = 17 missing);median (IQR)		65 (55–72)		-0.194	< **0.001**^{rs}
Gender (N = 11 missing); frequency (%)					0.872^{U}
Male	131 (27.8)		3 (3–4)		
Female	340 (72.2)		3 (3–4)		
Deprivation index (N = 85 missing);median (IQR)		4(2–7)		-0.084	0.098^{rs}
Employment (N = 6 missing); frequency (%)					**0.001**^{H}
Employed	146 (30.7)		3 (3–4)		
Unemployed	86 (18.1)		3 (3–4)		
Retired	240 (50.4)		3 (2–4)		
Other	4 (0.8)		4 (4–4)		
Ethnic group (N = 3 missing); frequency (%)					0.260^{H}
White	406 (84.8)		3 (3–4)		
Mixed	9 (1.9)		3 (3–4)		
Asian	37 (7.7)		3 (3–4)		
Black	26 (5.4)		3 (3–4)		
Other	1 (0.2)		4 (4–4)		
Smoking (N = 9 missing); frequency (%)					0.200^{H}
Current	53 (11.2)		3 (3–4)		
Ever	158 (33.4)		3 (3–4)		
Never	262 (55.4)		3 (2–4)		
Education (N = 35 missing); frequency (%)					0.356^{U}
A level or lower	300 (67.1)		3 (3–4)		
Higher than A level	147 (32.9)		3 (3–4)		
Current treatment; frequency (%)					
No treatment	11 (2.3)		3 (3–3)		0.524^{U}
Conventional synthetic DMARDs and glucocorticoids	428 (89.4)		3 (3–4)		0.568^{U}
Biologic DMARDs	156 (32.6)		3 (3–4)		0.690^{U}
RA duration (years) (N = 97 missing); median (IQR)		10 (4–20)		-0.175	**0.001**^{rs}
RAID score (N = 15 missing); median (IQR)					
Pain ($N = 2$ missing)		5(3–7)		0.030	0.515^{rs}
Ability ($N = 4$ missing)		5 (2–7)		-0.013	0.774^{rs}
Fatigue ($N = 7$ missing)		6 (3–8)		0.050	0.279^{rs}
Sleep ($N = 3$ missing)		5 (2–7)		0.026	0.576^{rs}
Physical wellbeing ($N = 4$ missing)		5 (3–7)		-0.002	0.965^{rs}
Emotional wellbeing ($N = 4$ missing)		4 (2–7)		-0.001	0.975^{rs}
Coping ($N = 4$ missing)		4 (2–6)		-0.019	0.681^{rs}
Brief illness perception questionnaire; median (IQR)					
Consequences ($N = 5$ missing)		6 (4–8)		0.017	0.716^{rs}
Timeline ($N = 15$ missing)		10 (9–10)		-0.016	0.732^{rs}
Personal control ($N = 7$ missing)		5 (4–7)		-0.044	0.344^{rs}
Treatment control ($N = 4$ missing)		8 (6–9)		-0.013	0.787^{rs}
Identity ($N = 3$ missing)		6 (5–8)		0.030	0.510^{rs}
Concern ($N = 7$ missing)		7 (5–9)		0.071	0.127^{rs}
Understanding ($N = 3$ missing)		8 (6–9)		0.044	0.340^{rs}
Emotional ($N = 7$ missing)		6 (3–8)		0.043	0.354^{rs}
Health literacy ($N = 7$ missing); median (IQR)		0 (0–1)		-0.045	0.334^{rs}
Health numeracy ($N = 8$ missing); median (IQR)		14 (11–17)		0.003	0.947^{rs}
Information Seeking ($N = 8$ missing); median (IQR)		84 (75–97)		0.261	< **0.001**^{rs}
Decision making ($N = 7$ missing); median (IQR)		54 (42–67)		0.092	**0.048**^{rs}
Brief Avoidance Coping Questionnaire ($N = 17$ missing); median (IQR)		28 (25–31)		0.085	0.069^{rs}
Optimism ($N = 5$ missing); median (IQR)		8 (6–9)		0.030	0.512^{rs}
Openness ($N = 1$ missing); median (IQR)		2 (1–3)		0.133	**0.004**^{rs}
Interest in predictive testing					
Children ($N = 65$ missing); median (IQR)		3 (2–3)		0.440	< **0.001**^{rs}
Siblings ($N = 87$ missing); median (IQR)		2 (2–3)		0.440	< **0.001**^{rs}
Family functioning ($N = 40$ missing); median (IQR)		2 (2–3)		0.226	< **0.001**^{rs}
Attitudes towards testing – median (IQR)					
Increased empowerment over person's health ($N = 9$ missing);		2.21 (1.98–2.45)		0.355	< **0.001**^{rs}

(*continued on next page*)

Table 1 (*continued*)

Patient characteristics	Descriptive statistics				
	Frequency for patients' characteristics	Medians (IQRs) for patients' characteristics	Medians (IQRs) for patients' likelihood of communicating risk	Test statistics	P value
Psychological harm as a result of knowing risk ($N = 8$ missing)		1.62 (1.37–1.99)		-0.155	**< 0.001**[rs]
Responsibility to obtain risk information ($N = 5$ missing)		-1.96 (−2.24 to 1.55)		-0.304	**< 0.001**[rs]
Social consequences as a result of predictive testing ($N = 4$ missing)		1.72 (0.87–2.17)		-0.032	0.483[rs]
Stress and avoidance around taking a predictive test ($N = 7$ missing)		1.25 (1.04–1.67)		-0.332	**< 0.001**[rs]

[rs]= Spearman's rank correlations, [H]= Kruskal-Wallis H test, [U]= Mann-Whitney U test. Correlation coefficients are reported for Spearman's rank correlations, medians and IQRs are reported for Kruskal-Wallis H and Mann-Whitney U tests. A positive Spearman's Rank correlation indicates that those with higher values for a patient characteristic were more likely to communicate about RA risk to their relatives. Significant variables ($p < 0.05$) are highlighted in bold.

Table 2
Response frequencies for patients' likelihood of communicating RA risk information to their relatives.

	Response frequencies (%) for patients' likelihood of communicating RA risk				
	Extremely unlikely	Unlikely	Neither likely nor unlikely	Likely	Extremely likely
All relatives (n = 1684)	**124 (7.4)**	**158 (9.4)**	**137 (8.1)**	**644 (38.2)**	**621 (36.9)**
Relationship of relative to the patient*					
Children (n = 792)	34 (4.3)	63 (8)	52 (6.5)	327 (41.3)	316 (39.9)
Siblings (n = 511)	63 (12.3)	56 (11)	38 (7.4)	167 (32.7)	187 (36.6)
Gender of relative*					
Male (n = 623)	44 (7)	64 (10.3)	43 (6.9)	231 (37.1)	241 (38.7)
Female (n = 680)	53 (7.8)	55 (8.1)	47 (6.9)	263 (38.7)	262 (38.5)

*Response frequencies for specific relatives (children vs siblings; male vs female) includes only those cases where patients indicated the characteristics of the relative (child, sibling, male, female) in relation to whom they were reporting their likelihood of communicating RA.

Table 3
Wilcoxon tests for reported relatives' characteristics and their association with patients' likelihood of communicating risk.

Reported relatives' characteristics	Response frequencies (%) for patients' likelihood of communicating RA risk*					Medians (IQRs)	P Value
	Extremely unlikely (0)	Unlikely (1)	Neutral (2)	Likely (3)	Extremely likely (4)		
Relationship to the patient							
Children	11 (5.8)	15 (7.9)	13 (6.9)	66 (34.7)	85 (44.7)	3.00 * * (3.00–4.00)	< 0.001
Siblings	25 (13.1)	24 (12.7)	18 (9.5)	58 (30.5)	65 (34.2)	3.00 (1.38–4.00)	
Gender							
Male	17 (7.7)	24 (10.9)	22 (10)	68 (30.8)	90 (40.7)	3.00 (2.00–4.00)	0.317
Female	15 (6.8)	19 (8.6)	21 (9.5)	76 (34.4)	90 (40.7)	3.00 (2.75–4.00)	

*Response frequencies relate to patients who reported having both male and female relatives, or both child and sibling relatives.
* * A median score of 3 indicates that a patient is likely to communicate RA risk information to their FDR.

their likelihood of communicating risk information was not significantly influenced by their FDRs' gender ($p = 0.317$) (Table 3).

Principle components analysis of the 23 items describing advantages and disadvantages of predictive testing was conducted. Factor loadings with an absolute value of < 0.3 were disregarded.[50] The Kaiser-Meyer-Olkin measure of sampling adequacy was 0.87. Bartlett's test of sphericity was significant ($p < 0.001$). A five-factor solution (Table 4) explained 63.16% of the variance. After interpretation of the factor loadings, the factors were labelled as: (i) Increased empowerment over person's health; (ii) Psychological harm as a result of knowing risk; (iii) Responsibility to obtain risk information; (iv) Social consequences as a result of predictive testing and (v) Stress and avoidance around taking a predictive test. From the univariate analyses, thirteen predictor variables were independently associated with patients' likelihood of communicating RA risk information to their FDRs (Table 1).

Following the backward elimination variable selection method, seven variables were included in the final multivariate regression. A flow chart detailing this process can be found in supplemental material 1. The final model is outlined in Table 5.

VIFs for these predictor variables were satisfactory, ranging from 1.08 to 1.32. Correlation coefficients among pairs of predictor variables were not large, ranging from − 0.31–0.52. Tables showing the VIFs and correlation coefficients for each of these variables can be found in supplemental material 2.

Patients were less likely to communicate RA risk information to their FDRs if they had stronger beliefs that tests to predict the risk of RA would cause stress to their relatives. Patients were more likely to communicate risk information to their FDRs if they had higher preferences for autonomy in health-related decision-making and stronger beliefs that risk knowledge would increase people's empowerment over their health. Patients who were 'not interested' in their children taking a predictive test for RA reported being less likely to communicate RA risk information compared to those who were 'definitely interested' in their children taking a predictive test. Those who were 'probably interested' in having their children take a predictive test were no more likely to communicate risk compared to those who were 'definitely interested'. Finally, patients who were 'not interested' or 'probably interested' in their siblings taking a predictive test for RA reported being less likely to communicate RA risk information compared to those who were 'definitely interested' in their siblings taking a predictive test.

Reasons patients were likely / unlikely to communicate RA risk information to their relatives are summarised in Table 6. The three most

Table 4
Factor labels and loadings from a factor analysis measuring patients' attitudes towards predictive testing.

Factors	Items	Factor loadings
1. Increased empowerment over person's health	"Finding out they were at high risk of developing RA would help a person feel prepared if they developed symptoms of RA"	0.848
	"Finding out their risk of developing RA would help a person to make important decisions about how to live their lives"	0.835
	"Finding out they were at high risk of developing RA would help a person get treated quickly if they developed symptoms of RA"	0.782
	"A person found to be at high risk of developing RA would be able to lower their risk by making changes to their lifestyle"	0.778
	"Finding out their risk of developing RA would give a person control over their health"	0.745
	"A person found to be at high risk of developing RA would be able to lower their risk by taking medications"	0.620
	"Knowing that their risk of developing RA was low would bring a person peace of mind"	0.550
2. Psychological harm as a result of being at high risk	"People found to be at high risk of developing RA may become anxious as a result"	0.918
	"People found to be at high risk of developing RA may become depressed as a result"	0.842
	"People found to be at high risk of developing RA are likely to worry unnecessarily about their health"	0.745
	"The relatives of someone found to be at high risk of developing RA would be upset"	0.651
	"Parents found to be at high risk of developing RA are likely to feel guilty about the about the possibility of passing the risk on to their children"	0.605
	"Knowing that they were at high risk of developing RA would harm a person's self-image"	0.449
3. Responsibility to obtain risk information	"People should find out their risk of developing RA to determine whether their children might be at risk"	-0.836
	"People should find out their risk of developing RA for the sake of their family"	-0.828
	"People should find out their risk of developing RA at an early age"	-0.765
	"Getting a test to predict their risk of developing RA would tell a person that they definitely would, or wouldn't develop RA"	-0.674
	"Not knowing their risk of developing RA could make a person anxious"	-0.623
4. Social consequences as a result of testing	"People found to be at high risk of developing RA may not be able to get insurance"	0.902
	"People found to be at high risk of developing RA may be discriminated against"	0.844
5. Stress and avoidance around taking a predictive test	"Getting a test to predict their risk of developing RA would be a stressful experience for a person"	0.639
	"I prefer not to think about things that might never happen"	0.623
	"Getting a test to predict the risk of a person developing RA would be a stressful experience for their relatives"	0.614

Table 5
Final ordinal logistic regression model to predict patients' likelihood of communicating RA risk to FDRs.

Patients' predictors	OR (95% CI)	P-Value
Age	1.01 (0.99–1.03)	0.334
Stress and avoidance around taking a predictive test	2.96 (1.79–4.91)	< 0.001
Increased empowerment over health	0.41 (0.23–0.71)	0.002
Decision-making preferences	0.99 (0.97–1.00)	0.047
Interest in children taking a predictive test (reference category- Definitely interested)		
Probably interested	0.93 (0.45–1.91)	0.845
Not interested	4.00 (1.44–11.12)	0.008
Interest in siblings taking a predictive test (reference category- Definitely interested)		
Probably interested	2.70 (1.30–5.58)	0.007
Not interested	3.12 (1.29–7.56)	0.012
Dispositional openness (reference category- strongly agree)		
Agree	0.44 (0.19–1.03)	0.059
Neither agree nor disagree	0.84 (0.32–2.17)	0.716
Disagree	0.93 (0.39–2.18)	0.865
Strongly disagree	0.88 (0.29–2.67)	0.825

n = 164/3856 missing cases. OR: odds ratio. The outcome variable (likelihood of communicating RA risk) is scored as: 0 (extremely likely to communicate RA risk), 1 (likely to communicate RA risk) and 2 (unlikely/unsure about communicating RA risk).

cited reasons that patients were unlikely to communicate RA risk information to their relatives include the fact that their relatives feel healthy at the present time (45.0%), that they do not want to worry their relatives (36.1%) and that their relatives have other problems to deal with (34.3%).

4. Discussion

This study is the first quantitative assessment of the likelihood of RA patients communicating RA risk information to their FDRs.

Patients reported willingness to communicate RA risk information. This is consistent with previous qualitative work. [20] Our findings also align with those of previous studies, with patients being more likely to communicate risk information to their children compared with their siblings.[23,24,26].

The finding that patients' likelihood of communicating RA risk was not significantly influenced by their FDRs' gender contradicts previous research examining risk communication for other chronic diseases.[13, 23] The majority of the previous studies, however, assessed family communication about risk for breast and ovarian cancer.

Various patient characteristics were associated with their likelihood of communicating RA risk information to their FDRs. These included greater preferences for autonomy in health-related decision-making, stronger beliefs that risk knowledge would increase a person's empowerment over their health, and higher interest in their FDRs taking a predictive test for RA.

The influence of patients' preferences for autonomy in health-related decision-making and beliefs that risk knowledge would increase a person's empowerment over their health aligns with previous studies examining other diseases, such as breast and ovarian cancer.[14,30] Those studies found that patients were motivated to communicate

Table 6
Response frequencies for reasons patients were unlikely to communicate RA risk information to their relatives.

Items	Response frequency (%)					
	Definitely does not apply	Does not apply	Neutral	Applies	Definitely applies	% Applies/definitely applies
Not concerned about RA risk						
"They feel healthy at the present time"	55 (16.8)	49 (15.0)	76 (23.2)	121 (37.0)	26 (8.0)	45.0
"I'm not worried about the possibility that they might develop RA"	62 (18.9)	56 (17.1)	138 (42.1)	52 (15.9)	20 (6.1)	22.0
"I think that their risk of developing RA is low"	49 (14.6)	57 (17)	161 (48.1)	50 (14.9)	18 (5.4)	20.3
Nothing will be done to lower risk						
"They would be unlikely to do anything about their risk of developing RA"	33 (10.1)	57 (17.4)	131 (40.1)	92 (28.1)	14 (4.3)	32.4
"There is nothing that can be done to lower their risk of developing RA"	52 (15.7)	52 (15.7)	138 (41.6)	67 (20.2)	23 (6.9)	27.1
Avoidance of risk knowledge						
"They would rather not think about the possibility that they might develop RA"	35 (10.7)	57 (17.4)	124 (37.9)	85 (26.0)	26 (8.0)	34.0
"I would rather not think about the possibility that they might develop RA"	57 (17.2)	60 (18.1)	118 (35.5)	69 (20.8)	28 (8.4)	29.2
"I don't like talking about my RA with them"	95 (29.3)	81 (25.0)	81 (25.0)	57 (17.6)	10 (3.1)	20.7
"It is not my responsibility"	86 (26.6)	79 (24.5)	116 (35.9)	37 (11.5)	5 (1.5)	13.0
"They don't like it when I talk about my RA"	109 (32.8)	105 (31.6)	89 (26.8)	23 (6.9)	6 (1.8)	8.7
"I don't want them to know that I've got RA"	165 (49.8)	108 (32.6)	47 (14.2)	9 (2.7)	2 (0.6)	3.3
Age						
"They are too old"	109 (34.2)	88 (27.6)	80 (25.1)	34 (10.7)	8 (2.5)	13.2
"They are too young"	115 (36.4)	86 (27.2)	75 (23.7)	29 (9.2)	11 (3.5)	12.7
Privacy issues						
"I would feel that I was invading their privacy"	98 (30.3)	99 (30.7)	82 (25.4)	39 (12.1)	5 (1.5)	13.6
"They would feel that I was invading their privacy"	100 (31.1)	100 (31.1)	87 (27)	31 (9.6)	4 (1.2)	10.8
Anxiety/guilt surrounding RA						
"I don't want to worry them"	53 (16.3)	54 (16.6)	101 (31.0)	96 (29.4)	22 (6.7)	36.1
"The conversation would make me feel anxious"	74 (23.2)	83 (26.0)	98 (30.7)	50 (15.7)	14 (4.4)	20.1
"I would feel guilty"	99 (30.7)	111 (34.4)	76 (23.5)	31 (9.6)	6 (1.9)	11.5
"They might feel embarrassed"	91 (28.3)	83 (25.9)	113 (35.2)	26 (8.1)	8 (2.5)	10.6
"They might blame me"	103 (32.0)	112 (34.8)	74 (23.0)	28 (8.7)	5 (1.6)	10.3
"I might feel embarrassed"	121 (37.2)	124 (38.2)	62 (19.1)	16 (4.9)	2 (0.6)	5.5
Other life issues						
"They have other problems to deal with"	63 (19.5)	50 (15.5)	99 (30.7)	87 (26.9)	24 (7.4)	34.3
"They are busy"	68 (21.2)	68 (21.2)	113 (35.2)	60 (18.7)	12 (3.7)	22.4
"I have other problems to deal with"	79 (24.9)	76 (24.0)	95 (30.0)	55 (17.4)	12 (3.8)	21.2
"I am busy"	89 (27.8)	97 (30.3)	110 (34.4)	18 (5.6)	6 (1.9)	7.5
Lack of knowledge surrounding RA						
"Doctors cannot tell them for certain that they will, or won't develop RA"	56 (16.7)	46 (13.7)	121 (36.0)	81 (24.1)	32 (9.5)	33.6
"They do not understand the impact that RA has on my life"	74 (22.8)	80 (24.6)	65 (20.0)	81 (24.9)	25 (7.7)	32.6
"I don't have enough information about their risk of developing RA"	67 (20.1)	58 (17.4)	104 (31.2)	77 (23.1)	27 (8.1)	31.2
"They think RA is something that affects older people"	90 (27.6)	83 (25.5)	86 (26.4)	55 (16.9)	12 (3.7)	20.6
Closeness with relatives						
"They live far away from me"	114 (35.5)	83 (25.9)	59 (18.4)	40 (12.5)	25 (7.8)	20.3
"I am not currently in contact with them"	141 (43.9)	83 (25.9)	46 (14.3)	29 (9.0)	22 (6.9)	15.9
"I do not have a close relationship with them"	132 (41.0)	91 (28.3)	56 (17.4)	22 (6.8)	21 (6.5)	13.3

Items shaded in grey indicate the ten items where participants responded with 'applies' or 'definitely applies' most frequently.

disease risk information to their relatives to empower them to obtain more information about their health and make important life decisions. [14,30] Predictive strategies for RA should therefore be developed in a way that facilitates autonomy and shared decision making.

Data from studies in CVD [34,35] are consistent with our finding that patients who had a higher interest in their FDRs taking a predictive test were more likely to communicate risk information to them. This reasoning may be associated with RA patients' beliefs that such tests would provide a high degree of certainty, and be able to rule in/ out RA development. [20,28].

Patients were more unlikely to communicate RA risk information to their FDRs if they believed that tests to predict the risk of RA would cause stress to a person and their relatives. This is consistent with previous qualitative studies highlighting concerns about stress and anxiety for relatives regarding their risk status [20] and underlines the importance of incorporating appropriate information and support to predictive and preventive strategies.

4.1. Implications

The findings from this study increase understanding of the process and determinants of communication about RA risk in families, and should inform the development of family risk information resources that are sensitive to patients' needs and concerns, and support patients and their FDRs to have an informed discussion. Further research is needed to explore patients' likelihood of communicating RA risk information to their FDRs through different channels (for example preferences for face to face, online or written communication).

4.2. Strengths and limitations

This paper has several methodological strengths, including a large sample size, the use of previously validated questionnaires, multidisciplinary contributors, and extensive patient partner involvement.

However, retired patients of white British origin were over-represented in the present sample and their views may not fully

represent those of other groups. This study was also limited to those within the West Midlands of the UK, and questionnaires were provided in English only. Further work is needed to capture the perspectives of diverse communities.

The sample for this study were self-selected and therefore may be subject to selection bias. As only those who returned their survey participated in the study, there were no data available for patients who did not respond to the survey. It would be informative to understand the characteristics and views of this non-participating group. Further work using alternative methodologies is needed to understand the views of FDRs who are unlikely to respond to a survey of this kind.

No objective measure assessed patients' disease activity in this study. Further investigation is needed to examine associations between patients' likelihood of communicating risk information to their FDRs, and measures of their disease activity including objective elements (e.g. DAS28).

Finally, the surveys provided to patients contained several questionnaires, which likely took considerable time to complete. Some questionnaires included within this survey (such as the brief IPQ and RAID) have been identified as complex to complete, which may increase patients' cognitive burden. [52–54] However, the survey was pre-tested by patient partners who felt that all items included within the patients' survey were relevant and that the survey was manageable.

5. Conclusion

Patients were willing to communicate RA risk to their FDRs and more likely to communicate about risk to their children than their siblings. Factors including decision-making preferences, interest in FDRs taking a predictive test, and beliefs that risk knowledge would increase a person's empowerment over their health increased the likelihood that patients would communicate RA risk information to FDRs. Beliefs that risk information would cause stress to their relatives reduced the likelihood that patients would communicate RA risk information. These findings are informative for the development of resources to support family communication about RA and RA risk, and facilitate access to FDRs to participate in risk reduction approaches or prediction/prevention studies.

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CRediT authorship contribution statement

IW contributed to data collection, data entry, data analysis and interpretation, and drafting of the manuscript. DZ contributed to data analysis and interpretation, and revision of the manuscript. GS contributed to study conception, design and revision of the manuscript. RJS contributed to study conception, design and revision of the manuscript. CDM contributed to revision of the manuscript. KR contributed to study conception, design, management, data analysis and interpretation, and revision of the manuscript. MF contributed to study conception, design, management, data collection, data analysis and interpretation, and revision of the manuscript.

Declaration of Competing Interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: IW, DZ, GS, RJS, KR and MF declare they have no competing interests. CDM is funded by the National Institute for Health Research (NIHR) Applied Research Collaboration West Midlands, the National Institute for Health Research (NIHR) School for Primary Care Research and a National Institute for Health Research (NIHR) Research Professorship in General Practice (NIHR-RP-2014–04-026) for this research project. The School of Medicine has received financial support from BMS for an unrelated non-pharmacological study screening patients for AF in primary care.

Data availability

The data underlying this article will be shared on reasonable request to the corresponding author.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.pec.2023.107713.

References

[1] Grassi W, De Angelis R, Lamanna G, Cervini C. The clinical features of rheumatoid arthritis. Eur J Radaiol 1998;27(Suppl 1):S18–24.
[2] van der Linden MP, le Cessie S, Raza K, van der Woude D, Knevel R, Huizinga TWJ. van der Helm-van Mil AHM. Long-term impact of delay in assessment of early arthritis patients. Arthritis Rheum 2010;62:3537–46.
[3] van Dongen H, van Aken J, Lard LR, Visser K, Ronday HK, Hulsmans HMJ, et al. Efficacy of methotrexate treatment in patients with probable rheumatoid arthritis: a double-blind, randomized, placebo-controlled trial. Arthritis Rheum 2007;56(5):1424–32.
[4] Gerlag DM, Safy M, Maijer KI, Tang MW, Tas SW, Starmans-Kool MJF, van Tubergen A, et al. Effects of B-cell directed therapy on the preclinical stage of rheumatoid arthritis: the PRAIRI study. Ann Rheum Dis 2019;78(2):179–85.
[5] Al-Laith M, Jasenecova M, Abraham S, Bosworth A, Bruce IN, Buckley CD, et al. Arthritis prevention in the pre-clinical phase of RA with abatacept (the APIPPRA study): a multi-centre, randomised, double-blind, parallel-group, placebo-controlled clinical trial protocol. Trials 2019;20(1):429.
[6] Frisell T, Holmqvist M, Källberg H, Klareskog L, Alfredsson L, Askling J. Familial risks and heritability of rheumatoid arthritis: role of rheumatoid factor/anti-citrullinated protein antibody status, number and type of affected relatives, sex, and age. Arthritis Rheum 2013;65(11):2773–82.
[7] Sugiyama D, Nishimura K, Tamaki K, Tsuji G, Nakazawa T, Morinobu A, et al. Impact of smoking as a risk factor for developing rheumatoid arthritis: a meta-analysis of observational studies. Ann Rheum Dis 2010;69(1):70–81.
[8] Feng J, Chen Q, Yu F, Wang Z, Chen S, Jin Z, et al. Body mass index and risk of rheumatoid arthritis: a meta-analysis of observational studies. Med (Baltim) 2016;95(8):e2859.
[9] Pattison DJ, Symmons DP, Lunt M, Welch A, Luben R, Bingham SA, et al. Dietary risk factors for the development of inflammatory polyarthritis: evidence for a role of high level of red meat consumption. Arthritis Rheum 2004;50(12):3804–12.
[10] Pre-clinical EValuation of Novel Targets in RA (PREVeNT-RA): a nationwide register of first-degree relatives of patients with rheumatoid arthritis to evaluate predictors of the development of RA. ⟨https://www.research.uhb.nhs.uk/trials/rrk4699/⟩. Accessed 22 Aug 2020.
[11] Arthritis-Checkup: study of an early detection of the disease. ⟨http://www.arthritis-checkup.ch/index_gb.html⟩ Accessed 22 Aug 2020.
[12] Kolfenbach JR, Deane KD, Derber LA, O'Donnell C, Weisman MH, Buckner JH, et al. A prospective approach to investigating the natural history of preclinical rheumatoid arthritis (RA) using first-degree relatives of probands with RA. Arthritis Rheum 2009;61(12):1735–42.
[13] Wiseman M, Dancyger C, Michie S. Communicating genetic risk information within families: a review. Fam Cancer 2010;9(4):691–703.
[14] Hallowell N, Ardern-Jones A, Eeles R, Foster C, Lucassen A, Moynihan C, et al. Communication about genetic testing in families of male BRCA1/2 carriers and non-carriers: patterns, priorities and problems. Clin Genet 2005;67(6):492–502.
[15] Koehly LM, Peters JA, Kenen R, Hoskins LM, Ersig AL, Kuhn NR, et al. Characteristics of health information gatherers, disseminators, and blockers within families at risk of hereditary cancer: implications for family health communication interventions. Am J Public Health 2009;99(12):2203–9.

[16] Shah LL, Daack-Hirsch S. Family communication about genetic risk of hereditary cardiomyopathies and arrhythmias: an integrative review. J Genet Couns 2018;27(5):1022–39.
[17] Simons G, Mallen CD, Kumar K, Stack RJ, Raza K. A qualitative investigation of the barriers to help-seeking among members of the public presented with symptoms of new-onset rheumatoid arthritis. J Rheuma 2015;42(4):585–92.
[18] Singhal J, Wells I, Simons G, Wöhlke S, Raza K, Falahee M. Public perceptions of predictive testing for rheumatoid arthritis compared to breast cancer and early-onset Alzheimer's disease: a qualitative study. BMC Rheuma 2022;6(1):14.
[19] Simons G, Belcher J, Morton C, Kumar K, Falahee M, Mallen CD, et al. Symptom recognition and perceived urgency of help-seeking for rheumatoid arthritis and other diseases in the general public: a mixed method approach. Arthritis Care Res 2017;69:633–41.
[20] Falahee M, Simons G, Buckley CD, Hansson M, Stack RJ, Raza K. Patients' perceptions of their relatives' risk of developing rheumatoid arthritis and of the potential for risk communication, prediction, and modulation. Arthritis Care Res (Hoboken) 2017;69(10):1558–65.
[21] Aletaha D, Neogi T, Silman AJ, Funovits J, Felson DT, Bingham CO, et al. rheumatoid arthritis classification criteria: an American College of Rheumatology/European League Against Rheumatism collaborative initiative. Ann Rheum Dis 2010 2010;69(9):1580–8.
[22] Healey E, Taylor N, Greening S, Wakefield CE, Warwick L, Williams R, et al. Quantifying family dissemination and identifying barriers to communication of risk information in Australian BRCA families. Genet Med 2017;19(12):1323–31.
[23] Young AL, Butow PN, Rhodes P, Tucker KM, Williams R, Healey E, et al. Talking across generations: family communication about BRCA1 and BRCA2 genetic cancer risk. J Genet Couns 2019;28(3):516–32.
[24] Stuttgen K, Bollinger J, McCague A, Dvoskin R, Mathews D. Family communication patterns and challenges of huntington's disease risk, the decision to pursue presymptomatic testing, and test results. J Huntingt Dis 2020;9(3):265–74.
[25] Haukkala A, Kujala E, Alha P, Salomaa V, Koskinen S, Swan H, et al. The return of unexpected research results in a biobank study and referral to health care for heritable long QT syndrome. Public Health Genom 2013;16(5):241–50.
[26] Forrest K, Simpson SA, Wilson BJ, van Teijlingen ER, McKee L, Haites N, et al. To tell or not to tell: barriers and facilitators in family communication about genetic risk. Clin Genet 2003;64(4):317–26.
[27] Wilson BJ, Forrest K, van Teijlingen ER, McKee L, Haites N, Matthews E, et al. Family communication about genetic risk: the little that is known. Community Genet 2004;7(1):15–24.
[28] Lea DH, Kaphingst KA, Bowen D, Lipkus I, Hadley DW. Communicating genetic and genomic information: health literacy and numeracy considerations. Public Health Genom 2011;14(4–5):279–89.
[29] Kaphingst KA, Blanchard M, Milam L, Pokharel M, Elrick A, Goodman MS. Relationships between health literacy and genomics-related knowledge, self-efficacy, perceived importance, and communication in a medically underserved population. J Health Commun 2016;21(Suppl 1):58–68.
[30] Metcalfe A, Coad J, Plumridge GM, Gill P, Farndon P. Family communication between children and their parents about inherited genetic conditions: a meta-synthesis of the research. Eur J Hum Genet 2008;16(10):1193–200.
[31] Klitzman R, Thorne D, Williamson J, Marder K. The roles of family members, health care workers, and others in decision-making processes about genetic testing among individuals at risk for Huntington disease. Genet Med 2007;9(6):358–71.
[32] Bachner YG, Carmel S. Open communication between caregivers and terminally ill cancer patients: the role of caregivers' characteristics and situational variables. Health Commun 2009;24(6):524–31.
[33] Munro H, Scott SE, King A, Grunfeld EA. Patterns and predictors of disclosure of a diagnosis of cancer. Psycho-Oncol 2015;24(5):508–14.
[34] Holt K. What do we tell the children? Contrasting the disclosure choices of two HD families regarding risk status and predictive genetic testing. J Genet Couns 2006;15(4):253–65.
[35] Batte B, Sheldon JP, Arscott P, Huismann DJ, Salberg L, Day SM, et al. Family communication in a population at risk for hypertrophic cardiomyopathy. J Genet Couns 2015;24(2):336–48.
[36] Chivers Seymour K, Addington-Hall J, Lucassen AM, Foster CL. What facilitates or impedes family communication following genetic testing for cancer risk? A systematic review and meta-synthesis of primary qualitative research. J Genet Couns 2010;19(4):330–42.
[37] Gossec L, Dougados M, Rincheval N, Balanescu A, Boumpas DT, Canadelo S. Elaboration of the preliminary Rheumatoid Arthritis Impact of Disease (RAID) score: a EULAR initiative. Ann Rheum Dis 2009;68(11):1680–5.
[38] Broadbent E, Petrie KJ, Main J, Weinman J. The brief illness perception questionnaire. J Psychosom Res 2006;60(6):631–7.
[39] Figueiras MJ, Alves NC. Lay perceptions of serious illnesses: an adapted version of the Revised Illness Perception Questionnaire (IPQ-R) for healthy people. Psychol Health 2007;22(2):143–58.
[40] Morris NS, MacLean CD, Chew LD, Littenberg B. The single item literacy screener: evaluation of a brief instrument to identify limited reading ability. BMC Fam Pr 2006;7(1):21.
[41] McNaughton CD, Cavanaugh KL, Kripalani S, Rothman RL, Wallston KA. Validation of a Short, 3-Item Version of the Subjective Numeracy Scale. Med Decis Mak 2015;35(8):932–6.
[42] Ende J, Kazis L, Ash A, Moskowitz MA. Measuring patients' desire for autonomy: decision making and information-seeking preferences among medical patients. J Gen Intern Med 1989;4(1):23–30.
[43] Finset A, Steine S, Haugli L, Steen E, Laerum E. The brief approach/avoidance coping questionnaire: development and validation. Psychol Health Med 2002;7(1):75–85.
[44] Scheier MF, Carver CS, Bridges MW. Distinguishing optimism from neuroticism (and trait anxiety, self-mastery, and self-esteem): a reevaluation of the Life Orientation Test. J Pers Soc Psychol 1994;67(6):1063.
[45] Figueiredo MI, Fries E, Ingram KM. The role of disclosure patterns and unsupportive social interactions in the well-being of breast cancer patients. Psycho-Oncol 2004;13(2):96–105.
[46] Epstein NB, Baldwin LM, Bishop DS. The McMaster family assessment device. J Marital Fam Ther 1983;9(2):171–80.
[47] Cameron LD, Sherman KA, Marteau TM, Brown PM. Impact of genetic risk information and type of disease on perceived risk, anticipated affect, and expected consequences of genetic tests. Health Psychol 2009;28(3):307.
[48] Stack RJ, Stoffer M, Englbrecht M, Mosor E, Falahee M, Simons G, et al. Perceptions of risk and predictive testing held by the first-degree relatives of patients with rheumatoid arthritis in England, Austria and Germany: a qualitative study. BMJ Open 2016;6(6):e010555.
[49] Falahee M, Simons G, Raza K, Stack RJ. Healthcare professionals' perceptions of risk in the context of genetic testing for the prediction of chronic disease: a qualitative metasynthesis. J Risk Res 2018;21(2):129–66.
[50] Bayliss K, Raza K, Simons G, Falahee M, Hansson M, Starling B, et al. Perceptions of predictive testing for those at risk of developing a chronic inflammatory disease: a meta-synthesis of qualitative studies. J Risk Res 2018;21(2):167–89.
[51] Wells I, Zemedikun DT, Simons G, Stack RJ, Mallen CD, Raza K, et al. Predictors of interest in predictive testing for rheumatoid arthritis among first degree relatives of rheumatoid arthritis patients. Rheumatol 2021;61(8):3223–33.
[52] Van Oort L., Schröder C., French D.P. What do people think about when they answer the Brief Illness Perception Questionnaire? A 'think-aloud' study. Br J Health Psychol 201; 16(2):231–245.
[53] ter Wee MM, van Tuyl LH, Blomjous BS, Lems WF, Boers M, Terwee CB. Content validity of the Dutch Rheumatoid Arthritis Impact of Disease (RAID) score: results of focus group discussions in established rheumatoid arthritis patients and comparison with the international classification of functioning, disability and health core set for rheumatoid arthritis. Arthritis Res Ther 2016;18(22):1–9.
[54] Lenzner T, Kaczmirek L, Lenzner A. Cognitive burden of survey questions and response times: a psycholinguistic experiment. Appl Cogn Psychol 2010;24(7):1003-2.