UNIVERSITY OF BIRMINGHAM
University of Birmingham Research at Birmingham

Does being exposed to an educational tool influence patient preferences?

Bywall, Karin; Veldwijk, Jorien; Hansson, Mats; baecklund, eva ; Raza, Karim; Falahee, M; Kihlbom, Ulrik

DOI:
10.1016/j.pec.2021.03.013

License:
Creative Commons: Attribution-NonCommercial-NoDerivs (CC BY-NC-ND)

Document Version
Peer reviewed version

Citation for published version (Harvard):
Bywall, K, Veldwijk, J, Hansson, M, baecklund, E, Raza, K, Falahee, M & Kihlbom, U 2021, 'Does being exposed to an educational tool influence patient preferences? The influence of an educational tool on patient preferences assessed by a discrete choice experiment.', *Patient Education and Counseling*, vol. 104, no. 10, pp. 2577-2585. https://doi.org/10.1016/j.pec.2021.03.013

Link to publication on Research at Birmingham portal

General rights
Unless a licence is specified above, all rights (including copyright and moral rights) in this document are retained by the authors and/or the copyright holders. The express permission of the copyright holder must be obtained for any use of this material other than for purposes permitted by law.

•Users may freely distribute the URL that is used to identify this publication.
•Users may download and/or print one copy of the publication from the University of Birmingham research portal for the purpose of private study or non-commercial research.
•User may use extracts from the document in line with the concept of 'fair dealing' under the Copyright, Designs and Patents Act 1988 (?)
•Users may not further distribute the material nor use it for the purposes of commercial gain.

Where a licence is displayed above, please note the terms and conditions of the licence govern your use of this document.

When citing, please reference the published version.

Take down policy
While the University of Birmingham exercises care and attention in making items available there are rare occasions when an item has been uploaded in error or has been deemed to be commercially or otherwise sensitive.

If you believe that this is the case for this document, please contact UBIRA@lists.bham.ac.uk providing details and we will remove access to the work immediately and investigate.

Download date: 18. Apr. 2024

Title: Does being exposed to an educational tool influence patient preferences?

Subtitle: The influence of an educational tool on patient preferences assessed by a discrete choice experiment.

Authors: Karin Schölin Bywall, Jorien Veldwijk, Mats G Hansson, Eva Baecklund, Karim Raza, Marie Falahee & Ulrik Kihlbom

Corresponding author at:

Karin Schölin Bywall

Department of Public Health and Caring Sciences, Centre for Research Ethics & Bioethics, Uppsala University, Uppsala, Sweden

karin.bywall@crb.uu.se

+46 184716249

Jorien Veldwijk, PhD

Erasmus School of Health Policy & Management

and Erasmus Choice Modelling Centre

Erasmus University Rotterdam

Rotterdam, the Netherlands

Department of Public Health and Caring Sciences, Centre for Research Ethics & Bioethics, Uppsala University, Uppsala, Sweden

veldwijk@eshpm.eur.nl

Mats Hansson, PhD

Department of Public Health and Caring Sciences, Centre for Research Ethics & Bioethics,

Uppsala University, Uppsala, Sweden

mats.hansson@crb.uu.se

Eva Baecklund, PhD

Department of Medical Sciences, Rheumatology

Uppsala University, Uppsala, Sweden

eva.baecklund@medsci.uu.se

Karim Raza, FRC PhD

Institute of Inflammation and Ageing

University of Birmingham Research Laboratories

Queen Elizabeth Hospital, Birmingham, UK

Sandwell and West Birmingham NHS Trust

Birmingham, UK

k.raza@bham.ac.uk

Marie Falahee, BSc PhD

Institute of Inflammation and Ageing

University of Birmingham Research Laboratories

Queen Elizabeth Hospital, Birmingham, UK

m.falahee@bham.ac.uk

Ulrik Kihlbom, PhD

Department of Public Health and Caring Sciences, Centre for Research Ethics & Bioethics,

Uppsala University, Uppsala, Sweden

ulrik.kihlbom@crb.uu.se

Highlights

- There is uncertainty regarding how patient preferences and the results of DCE studies are influenced by educational tools.
- Guidance on how to design and frame training materials for preference assessments is currently lacking.
- This study shows that using an educational tool to inform patients led to patients placing more importance on treatment side effects in their decision making.
- Patients receiving information via written text alone placed relatively more importance on treatment effectiveness and administration methods.
- Further research is needed to provide guidance on the making and use of educational tools in preference elicitation studies.

Abstract

Objectives: There is an increased interest in patient preferences informing the development and authorisation of medical products. A requirement for robust and meaningful results of such studies is that patients adequately understand the risks and benefits associated with treatments for which their preferences are elicited. This study aims to determine the influence of an educational tool, compared with traditional written information on patient preferences elicited in a discrete choice experiment (DCE).

Methods: Treatment preferences of Swedish patients with rheumatoid arthritis (RA) were assessed using a DCE. Patients were recruited via clinics, a research panel, and the Swedish Rheumatism Association. Respondents received training materials either as plain written text or as an online educational tool. The educational tool was designed to enhance understanding of the written text by using graphics, pictograms, icon arrays, spoken text, and click-on functions. Data were analysed using random parameter logit models.

Results: 675 patients with RA were included in the analysis. The patients received either a written information (n=358) or information via an educational tool (n=317). Respondents receiving the educational tool placed relatively more importance on all included side effects in their decision making, compared to respondents receiving the written text, who placed greater importance on treatment effectiveness and administration methods.

Conclusion: Compared to the respondents receiving the written text, the decisions of respondents receiving the educational tool were more influenced by medication side effects. Further research is needed to provide guidance on how and when to use educational tools to inform and elicit patients' preferences.

Practice implications: The ways in which attributes are presented to patients significantly impacts preferences measured in a DCE.

Keywords: Digital educational tools, Discrete choice experiments, Medical product lifecycle, Regulatory decisions, Rheumatoid arthritis

Funding: Financial support for this study was provided by the Patient Preferences in Benefit-Risk Assessments during the Drug Life Cycle (PREFER) is a five-year project that has received funding from the Innovative Medicines Initiative 2 Joint Undertaking under grant agreement No 115966. This Joint Undertaking receives support from the European Union's Horizon 2020 research and innovation programme and the European Federation of Pharmaceutical Industries and Associations (EFPIA). The funding agreement ensured the authors' independence in designing the study, interpreting the data, writing, and publishing the report.

The following author KR is supported by the Birmingham NIHR Biomedical Research Centre and is a member of the Research into Inflammatory Arthritis Centre Versus Arthritis and the MRC Versus Arthritis Centre for Musculoskeletal Ageing Research.

Acknowledgements: This text and its contents reflect the PREFER project's view and not the view of IMI, the European Union, or EFPIA. The authors would like to thank The Swedish Rheumatism Association for their support in the recruitment of patient research partners and participants in the survey. Our sincere gratitude to Marie Almquist and Marie Heidenvall for providing their expertise as patient research partners throughout the research. We also thank Anna Vikerfors from the Swedish Medical Product Agency and members in the PREFER project for reviewing the research process and results.

Appendix: Supplementary material

1. Introduction

Increased interest in the use of patient preferences to inform decision-making throughout the life cycle of medical products has raised several methodological questions and concerns among stakeholders [1]. Representatives from the medical community, patient organisations, pharmaceutical companies, regulatory authorisation agencies, and health technology assessment bodies have expressed concern regarding the current level of patients' understanding of treatment-related risks and benefits when responding to a patient preference study [2].

Patient preferences can be assessed using a discrete choice experiment (DCE), which assumes that people make rational choices based on perceived utility [3]. Utility is estimated by modelling the respondents' choices (trade-offs) between competing alternatives (attribute levels) that are described in hypothetical choice questions [4]. The attributes in a patient preference study often include levels of risks and their probabilities, information that may be difficult for the respondents to understand [5]. Communicating risk information is difficult as people often struggle to read, retain, and understand the information [6]. For example, respondents in DCEs may fail to understand the trade-offs they make when answering choice questions if they have not properly understood the risks and benefits associated with the treatment options.

Typically, respondents in a patient preference study receive some form of written training material before answering the choice questions. The training materials for medical products are intended to help respondents understand the risks and benefits associated with medical products so they can make informed decisions [7]. However, guidance on how to design and frame such training materials for preference assessments is lacking, and training materials are rarely presented in published articles [5].

Digital educational tools to train and motivate respondents have supported learning outcomes in previous assessments of patient preferences [8, 9]. Randomized controlled trials have demonstrated that educational tools can be used to improve patients' knowledge, decrease decisional conflict, and, in some cases, improve patient participation in shared decision-making [10]. There is a growing trend towards using patient preferences to inform decisions about regulatory marketing authorisation [11]. Furthermore, patient preference information gathered from a DCE may inform risk and benefit profiles used to gain approval for new medical products [12].

Educational tools can increase the quality of preference data by improving learning outcomes through a stimulating educational environment [13]. In addition, educational tools may help respondents evaluate the risks and benefits and the attribute levels (i.e. competing alternatives) included in a patient preference study. Moreover, educational tools may help respondents evaluate the benefits and side effects in a way that mirrors a real-life situation [14]. Responses to a preference study using an educational might be more based on an accurate understanding of a disease's impact on daily function, treatment choices, and their benefit/risk profiles [7]. However, it is still not clear to what extent educational tools influence patients' preferences. To this end, this study aims to determine the influence of an educational tool, compared with traditional written information on patient preferences elicited in a DCE.

2. Methods

2.1 Case study: Recruitment and respondents

A DCE survey was designed to assess treatment preference regarding treatment with biologics and Janus kinase inhibitors (JAK) inhibitors, and heterogeneity within these preferences, for patients with rheumatoid arthritis (RA) living in Sweden. The educational tool was designed to help the respondents evaluate the risks and benefits associated with hypothetical treatment options for second-line treatment for RA.

Recruitment of patients with RA started in November 2018 and ended in October 2019. In total, 675 patients with RA were included in the analysis. Patients were recruited via three sources: a research panel (n=162) (dynata.com); the Swedish Rheumatism Association (n=228); and the rheumatology clinic at Uppsala University Hospital, Sweden (n=285). The Swedish Rheumatism Association and the research panel distributed the invitation as an e-mail with a web link to the survey. The clinic distributed the invitation as a letter via post. The invitation included a request to participate, a web page link to the survey or a hard copy. The following inclusion criteria were used: RA diagnosis; between 18–80 years old; able to understand and answer the questions; and able to read and understand Swedish without aid. The survey was approved by the regional ethics review board in Uppsala, Sweden (Reg no. 2018/156). Data generation, storage, and sharing were governed by the General Data Protection Regulation (GDPR) Act and Uppsala University data protection and security policies. Informed consent was collected from all respondents.

2.2 Discrete choice experiment (DCE)

DCEs is a quantitative method used to assess preferences presented in a choice-based manner. The method provides hypothetical choice questions that include different attributes and attribute levels (i.e. competing alternatives) [15]. The attributes and attribute levels for the DCE were identified in a stepwise manner following established methodological standards [16] via a literature overview, validation meetings with experts, and focus groups with patients. A preliminary list of attributes and attribute levels drafted from the literature overview was revised based on feedback from the validation meetings and the focus group meetings. Seven attributes were included in the DCE: route of administration, frequency of use, probability of mild short-term side effects, probability of side effects changing appearance, probability of psychological side effects, probability of severe side effects, and treatment effectiveness. All attributes and attribute levels are listed in Figure 1.

Attribute	Level 1	Level 2	Level 3
Route of administration	Tablet	Injection	Drip
Frequency of use	Daily	Weekly	Monthly
Probability of mild short-term side effects nausea, vomiting or headache	Common 1 in 10	Uncommon 1 in 100	Rare 1 in 1000
Probability of side effects affecting appearance hair loss, weight changes or skin rash	Common 1 in 10	Uncommon 1 in 100	Rare 1 in 1000
Probability of psychological side effects anxiety, mood changes, depression or sleep disturbance	Common 1 in 10	Uncommon 1 in 100	Rare 1 in 1000
Probability of severe side effects that requires hospitalisation such as severe infections or allergic reactions	Common 1 in 10	Uncommon 1 in 100	Rare 1 in 1000
Effectiveness The ability to decrease inflammation and swelling of the joints, also pain and other symptoms.	30% Improvement	50% Improvement	70% Improvement

Figure 1. Attributes and attribute levels

2.3 Development of the educational tool

Respondents received the same training content in one of two forms: either as a written (plain) text or as an educational tool that used graphics, pictograms, icon arrays, voice-over, and click-on functions. In addition, the respondents recruited via the research panel were only receiving the information in written. The training material and the educational tool were developed in parallel with the DCE questions. First, an overview of the literature describing disease antirheumatic drugs (DMARDs) was drafted that described the disease context of RA and the attributes and attribute levels included in the DCE. The draft was reviewed by: two rheumatologists, two patient research partners, and the team of researchers. The patient research partners helped refine the language to fit the Swedish RA population. This team of researchers met a second time to review the changes made in response to the first round of comments.

The educational tool was developed and illustrated with assistance from MindBytes, a Belgian company that applies a theory-driven and evidence-based approach to developing interactive gamified educational tools (http://www.mindbytes.be). The (written) content of the educational tool was designed by the research team, and MindBytes developed a digital version using graphics, voice over, and click functions. This version allowed the respondents to move through the information however they wanted. A high level of realism and a moderate interactivity level were chosen on the basis of the study population's information needs assesses by input from the two rheumatologists and the patient research partners. Both the plain (written text) and enhanced versions (educational tool) contained the same information about RA, and the treatment attributes and attribute levels included in the DCE. We provided the respondents the following decision-making scenario: 'Think of a situation where your treatment is not working, your joints are swollen, you have pain or unbearable side effects, and you need to change to a second-line treatment'. The attribute levels for the

educational tool were given as illustrations, written text, and audio/spoken words. Pictographs and icon arrays were used to describe risks and percentages. A pilot test of the DCE and the educational tool was conducted in a sample of patients with RA (N=22). After the pilot test, the framing of RA and the description of the attribute levels were slightly changed based on the feedback on the study materials elicited from pilot study respondents.

2.4 Design

The DCE experimental design generated by NGene 1.0 (ChoiceMetrics, 2011), asked each respondent to answer 15 hypothetical choice questions with two alternatives that were characterised by varying attribute levels. The DCE was a forced choice experiment with no opt-out provision. Based on the data retrieved in the pilot test, a multinomial logit (MNL) model was fitted. Beta estimates were used to assign priors for the final experimental DCE design, which is a *d*-efficient (Bayesian) design [16]. The final design included 60 choice questions divided into four blocks. All of the attributes were displayed in each choice question. An overlap of three attributes was applied to reduce the cognitive burden to the respondents, i.e. three attributes were identical.

2.5 Statistical analysis

We observed the respondents' time spent on the training material and the DCE, their trading behaviour, perceptions of the information provided, and difficulty answering the choice questions. We also observed the consistency (scale parameter) in the choice data of the DCE [17]. Lighthouse Studios (9.5.3) were used to administrate the survey. SPSS® Statistics 20 and Nlogit® were used to analyse the results. All results were considered statistically significant when $p < 0.05$.

2.5.1 Demographic characteristics

Questions regarding the characteristics of patients were included in the survey to allow description of the study sample. These included demographic characteristics such as age, gender, educational level, and occupational status. In addition, the respondents were asked about the duration of RA, their experience of side effects of treatment for RA, time to onset of drug effect for their RA, and experience with DMARD treatment. Measures of health literacy [18] and subjective numeracy [19] were also included. Questions regarding the respondents' understanding of the background information and the difficulty of the questionnaire were also included to determine whether there were any differences in choice-making ability due to the training materials.

2.5.2 Attribute estimates and the role of the scale parameter

The patients' preferences were determined by attribute level estimates and the relative importance of the attributes. The attribute levels were estimated using a multinomial logit (MNL) model [20] as both preference heterogeneity and scale heterogeneity can influence parameter values obtained in a DCE. The scale parameter test by Swait and Louviere were used to assess whether there were any differences in the level of error variance (choice consistency) in the two data sets (i.e. those who used the educational tool and those who used the plain text [20].

2.5.3 Preference heterogeneity

The random parameter logit (RPL) model was used to estimate attribute levels and heterogeneity in preferences. Estimates of random parameters are considered to be an average value associated with a standard deviation (SD) describing the heterogeneity of preferences within the sample. Estimates of attribute level indicate the preference for a specific attribute level (i.e. the more preferred outcome has a higher estimate). The RPL model included all the

attribute levels for the choice questions presented in Table 4. The levels for all categorical attributes (other than effectiveness) were dummy coded. All the attribute levels were tested for interaction with the educational tool. The RPL parameter estimates for the interaction terms can be interpreted as preference-weight adjustments that apply only to respondents in the corresponding subgroup (educational tool).

2.5.4 Relative importance

Relative importance scores were calculated based on results of the RPL separately for the written text and the educational tool. The difference between the highest and lowest estimates of the attribute level was calculated for each attribute. The largest difference value was assigned a 1, representing the most important attribute. The other difference values were divided by the largest difference value, revealing the relative distance between all other attributes and the most important attribute.

2.5.5 Sensitivity analysis

As a first step, all the demographic characteristics were compared in the two data sets (plain text and educational tool) to explore if there were any statistically significant differences using χ^2 test within the sets that could influence the results and lead to preference heterogeneity. Next, the relative importance of attribute levels was estimated using the RPL model and compared (separately) within the demographic characteristics that were statistically different (gender and time to onset of drug effect) within the data sets. This approach was used to explore if there were any differences in treatment preferences. Gender and time to onset of drug effect were also tested (separately) as interactions with the attribute estimates in the RPL model to explore if these characteristics contributed to the respondents' preferences for attributes.

Additional analyses were performed using χ^2 test to explore if there were statistically significant differences in the demographics depending on the recruitment of respondents as three different sources were used for recruitment (i.e. the clinic, the research panel, and the Swedish Rheumatism Association). A secondary analysis using the RPL model was performed on the data file of respondents from the research panel to estimate the relative importance of attribute level estimates of female and male respondents.

Further analyses were performed using the RPL model to explore if there were differences in treatment preferences depending on how the respondents were recruited. The relative importance of the three data sets (i.e. the clinic, the research panel, and the Swedish Rheumatism Association) was compared to explore differences in treatment preferences. The recruitment source was also tested (separately for clinic, panel and association) as interactions of the attribute level estimates in the RPL model to explore if the recruitment method contributed to the respondents' preferences for attribute levels.

Differences in treatment preferences were also estimated based on the time spent on the training material to explore if the time spent on the survey influenced patient preferences. The relative importance of the attribute level estimates of the MNL model explored if respondents who spent less time (80%) on the training material had different treatment preferences than respondents on average.

3. Results

3.1 Respondents and demographic characteristics

A total of 675 patients with RA were included in the analysis, i.e., respondents receiving the training in plain text (n=358) and the educational tool (n=317). The text version was answered (i.e. at least 80% of the choice questions) by n=422. Respondents were removed for flat-lining, i.e., only choosing option A or B in 80% of the choice questions (n=29) and for answering the survey under 5 minutes (n=35). The educational tool version was answered (i.e. at least 80% of the choice questions) by n=333. Respondents were removed for flat-lining (n=3) and for answering the survey under 5 minutes (n=8).

The majority of the respondents were female (80%) and highly educated (47%). The same percentage worked full time (43%) as were retired or unemployed (43%). Most of the respondents reported a sufficient health literacy (57%) and medium numeracy (50%). The following three treatments were the most commonly taken by respondents for their RA: synthetic DMARDs only (56%), biologic DMARDs (34%), and JAK inhibitors (4%). Only 23% of the respondents had no experience with treatment side effects (Table 1.). Statistically significant differences between the two samples (i.e., educational tool and plain text) were seen in gender and time to onset of drug effect.

Table 1. Patient characteristics

		N total (%)	N Educational tool (%)	N Plain text (%)
Total		675 (100)	317 (100)	358 (100)
Gender				
	Female	539 (80)	267 (84)	272 (77)
	Male	133 (20)	50 (16)	83 (23)
Age groups (years)				
	18–24	24 (4)	9 (3)	15 (4)
	25–34	59 (9)	17 (5)	42 (12)
	35–44	63 (9)	32 (10)	31 (9)
	45–54	131 (19)	67 (21)	64 (18)
	55–64	182 (27)	83 (26)	99 (28)
	65–80	214 (32)	109 (34)	105 (30)
Education level				
	Low	208 (31)	103 (32)	105 (30)
	Medium	151 (22)	62 (19)	89 (25)
	High	314 (47)	152 (48)	162 (45)
Occupational status				
	Full time employee, part time employee, parental leave/occupational leave	290 (43)	136 (43)	154 (43)
	Work part time since RA, long term sick leave, sick pension	140 (21)	61 (19)	79 (22)
	Age pensioner /unemployed	290 (43)	113 (36)	177 (33)
	Other	13 (2)	7 (2)	6 (2)
Health literacy				
	Sufficient	382 (57)	185 (58)	197 (55)
	Problematic	249 (37)	115 (36)	134 (38)
	Lacking	29 (4)	15 (5)	24 (7)
Numeracy				
	High	41 (6)	13 (4)	28 (8)
	Medium	335 (50)	214 (68)	212 (60)
	Low	201 (30)	88 (28)	113 (32)
Disease duration				
	1–12 months	44 (7)	22 (7)	22 (6)
	1–5 years	163 (24)	75 (24)	88 (25)
	5–10 years	119 (18)	52 (16)	67 (19)
	More than 10 years	347 (51)	168 (53)	179 (50)
Time to onset of drug effect				
	0–3 months	225 (33)	104 (33)	121 (34)
	3–12 months	163 (24)	76 (24)	87 (25)
	1–2 years	61 (9)	28 (9)	33 (9)
	2–5 years	75 (11)	38 (12)	37 (11)
	More than 5 years	82 (12)	50 (16)	32 (9)
	Still not working	64 (9)	21 (7)	43 (12)
Experience with treatment				
	First line treatment only (synthetics)	375 (56)	193 (61)	182 (51)
	Second line treatment			
	Biologics	231 (34)	115 (36)	116 (32)
	JAK inhibitors	26 (4)	14 (4)	12 (3)
Experience with side effects				
	Mild short term	405 (60)	200 (63)	205 (57)
	Appearance	307 (45)	153 (48)	154 (43)
	Psychological	234 (35)	97 (31)	137 (38)
	Severe	136 (20)	56 (18)	80 (22)
	No side effects	158 (23)	69 (22)	89 (24)

The average time for completing the full survey was 18 minutes for respondents using the plain text and 27 minutes for respondents using the educational tool. On average, respondents who used the plain text spent two minutes on the training material and respondents who used the educational tool spent nine and a half minutes on the training material, Most of the respondents perceived the training material as enough information (78%). Only a small percentage of the respondents (3–4%) reported difficulty answering the choice questions (Table 2).

Table 2. Survey characteristics

		Educational tool	Plain text
Time spend on survey			
	Average time	27 min	18 min
	Average time spend on training material	9.½ min	2 min
	Average time spend on DCE	17.½ min	16 min
How did you perceive the information provided?			
		Mean (%)	Mean (%)
	Too much information	47 (15)	66 (19)
	Enough information	249 (79)	276 (78)
	Too little information	3 (1)	8 (2)
How did you perceive answering the choice question?			
	It was easy for me to answer	120 (38)	136 (38)
	I had to think a bit before I could answer	168 (53)	202 (57)
	I had a hard time answering	12(4)	6 (3)

3.2 Preference heterogeneity

All attributes significantly influenced decision making, most showing significant SD for heterogeneity. A strong impact on patients' preferences depending on the received training material was revealed when including the educational tool as an interaction (Table 3).

Table 3. Preferences of patients based on random parameter logit model (pooled data file)

		Estimate	SE	SD	SE (SD)
Route of administration					
	Tablet	1.33***	0.09	0.79***	0.10
	Injection	0.58***	0.07	0.80***	0.07
	Drip (ref)				
Frequency of use					
	1 a day	−0.89***	0.07	0.59***	0.09
	1 a week	−0.40***	0.06	0.10	0.08
	1 a month (ref)				
Probability of mild short-term side effects					
	1 in 10	−0.51***	0.07	0.02	0.17
	1 in 100	−0.26***	0.05	0.32***	0.08
	1 in 1000 (ref)				
Probability of side effects changing appearance					
	1 in 10	−1.00***	0.07	0.06	0.14
	1 in 100	−0.13*	0.06	0.11	0.13
	1 in 1000 (ref)				
Probability of psychological side effects					
	1 in 10	−1.90***	0.09	1.05***	0.08
	1 in 100	−0.48***	0.07	0.08	0.17
	1 in 1000 (ref)				
Probability of severe side effects					
	1 in 10	−2.50***	0.10	1.34***	0.10
	1 in 100	−0.69***	0.06	0.06	0.10
	1 in 1000 (ref)				
Effectiveness (linear)		0.07***	0.00	0.04***	0.00
Interactions with training material (educational tool)					
Route of administration					
	Tablet	−0.05	0.17		
	Injection	−0.25**	0.12		
	Drip (ref)				
Frequency of use					
	1 a day	1.09***	0.20		
	1 a week	0.45***	0.11		
	1 a month (ref)				
Probability of mild short-term side effects					
	1 in 10	0.36***	0.13		
	1 in 100	−0.58***	0.12		
	1 in 1000 (ref)				
Probability of side effects changing appearance					
	1 in 10	−0.26**	0.11		
	1 in 100	−0.32**	0.14		
	1 in 1000 (ref)				
Probability of psychological side effects					
	1 in 10	0.57***	0.14		
	1 in 100	0.38***	0.13		
	1 in 1000 (ref)				
Probability of severe side effects					
	1 in 10	0.24**	0.12		
	1 in 100	0.08	0.13		
	1 in 1000 (ref)				
Effectiveness (linear)		−0.00***	0.00		

3.3 Relative importance

The outcome of the scale test showed that the estimates, but not the choice consistency, were significantly different across the two data sets (Table 4). The relative importance score revealed that the probability of a severe side effect compared to treatment effectiveness was relatively more important for respondents receiving the educational tool than for respondents receiving the plain text. Both groups had similar preferences for third place (probability of getting a psychological side effect), fourth place (route of administration), and fifth place (probability of getting a side effect affecting appearance). Least important were frequency of use and the probability of mild short-term side effects. Respondents receiving the educational tool were more influenced by the probabilities of all side effects than respondents receiving the plain text (Figure 2).

Table 4. Preferences of patients based on random parameter logit model

Attribute		Plain text				Education tool			
		Estimate	(SE)	SD	SE (SD)	Estimate	(SE)	SD	SE (SD)
Route of administration									
	Tablet	1.45***	0.11	0.91***	0.13	1.12***	0.11	0.70***	0.15
	Injection	0.73***	0.09	0.86***	0.11	0.44***	0.08	0.69***	0.10
	Drip (ref)								
Frequency of use									
	1 a day	−1.04***	0.08	0.60***	0.11	−0.28***	0.09	0.84***	0.12
	1 a week	−0.39***	0.07	0.10	0.12	−0.13	0.08	0.04	0.15
	1 a month (ref)								
Probability of mild short-term side effects									
	1 in 10	−0.46***	0.08	0.05	0.28	−0.73***	0.09	0.01	0.18
	1 in 100	−0.07	0.06	0.35***	0.11	−0.78***	0.07	0.27	0.18
	1 in 1000 (ref)								
Probability of side effects changing appearance									
	1 in 10	−1.02***	0.08	0.13	0.15	−1.13***	0.09	0.07	0.18
	1 in 100	−0.22**	0.08	0.07	0.24	−0.08	0.08	0.22	0.16
	1 in 1000 (ref)								
Probability of psychological side effects									
	1 in 10	−1.63***	0.11	1.05***	0.10	−2.02***	0.12	0.88***	0.12
	1 in 100	−0.43***	0.08	0.18	0.28	−0.57***	0.09	0.01	0.15
	1 in 1000 (ref)								
Probability of severe side effects									
	1 in 10	−2.18***	0.12	1.12***	0.13	−2.58***	0.14	1.34***	0.14
	1 in 100	−0.58***	0.07	0.07	0.12	−0.68***	0.08	0.06	0.19
	1 in 1000 (ref)								
Effectiveness (linear)		0.07***	0.00	0.05***	0.00	0.06***	0.00	0.03***	0.00

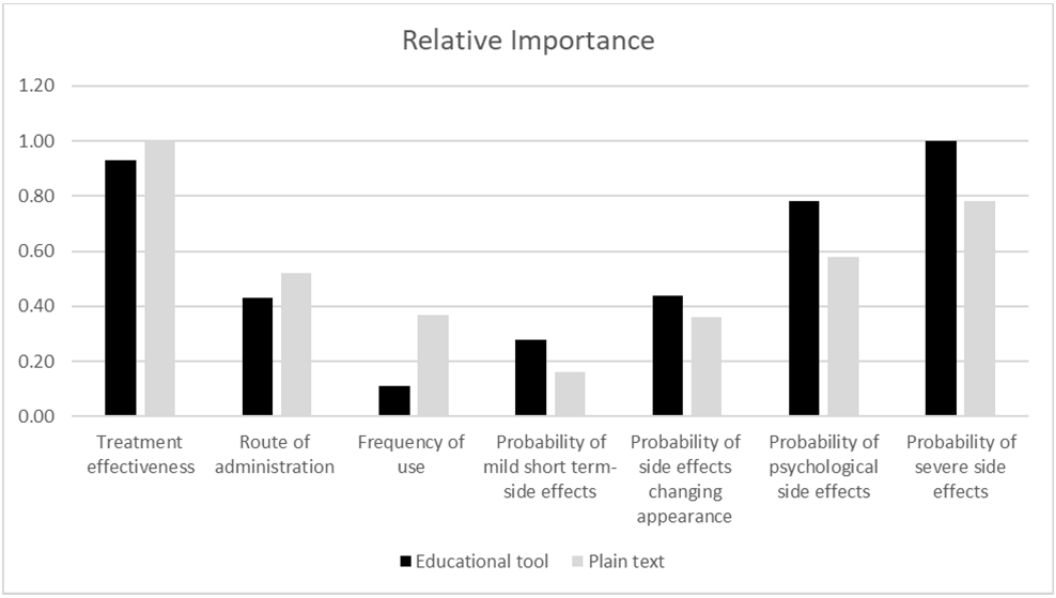


Figure 2. Relative importance

3.4 Sensitivity analysis

The sensitivity analysis revealed statistically significant differences in the two data sets (i.e. plain text and the educational tool) in gender and time to onset of drug effect. Respondents of the plain text version included more males and more responses indicating treatment was not working. However, gender or time to onset of drug effect did not contribute to the respondents' preferences.

A statistically significant difference for gender was also identified in respondents recruited by the research panel and this group included more males than the other recruitment sources. The RPL model revealed that gender did not statistically contribute to the preferences of the respondents from the research panel. Respondents' preferences for attribute levels were not statistically influenced by the recruitment source (i.e. respondents recruited from the clinic, the research panel, and the Rheumatism Association).

The relative importance of the attribute level estimates of the MNL model did not identify any differences in respondents who spent less time (80%) on the training material than respondents on average.

4. Discussion and Conclusion

4.1 Discussion

This study aimed to determine the influence of an educational tool, compared with traditional written information on patient preferences elicited in a DCE. This study revealed that respondents' preferences differed depending on whether they received the training material in the form of a plain text or in the form of an educational tool using graphics, pictograms, icon arrays, spoken text, and click-on functions. Compared to respondents receiving the same information in plain text, respondents receiving the educational tool put relatively more importance on all the probabilities of a side effect. In contrast to the results of this study, previous research has found that training materials in the form of a web-based interactive animated storyline about a disease and an intervention positively influenced the quality of choice data collected in a DCE in terms of the choice consistency [7]. Another study has found that enhanced animated information may not improve respondents' knowledge when compared to well-designed static training materials [21]. That is, animated training materials may be less impactful communicating information on risks and benefits than a simple version of an educational tool such as the one presented here (i.e., using graphics, pictograms, icon arrays, spoken text, and click-on functions).

As most people struggle to understand information on risks and benefits associated with medical treatment [6, 22], using an educational tool might stimulate participants in a patient preference study to further reflect on the risks and benefits of the treatment in the choice tasks. Further reflection and engagement makes objects less distal and more concrete, an understanding that influences preferences [23]. Respondents in this study spent more time on the training material if they received the educational tool rather than the plain text (Table 2). According to construal level theory, people's preferences for an object depend on their psychological distance from the object [24]. Psychological distance is a multi-dimensional

construct comprised of spatial distance, social distance, temporal distance, and hypotheticality. Temporal distance (how close or distant the event is in time) and hypotheticality (how imagined or real – i.e. how unlikely or likely the event is) are the most relevant dimensions for health care preferences in a preference study. The educational tool in our study may have allowed the participants to primarily reduce the hypotheticality of the risk attributes because they were experienced more vividly and thereby assigned greater importance.

A limitation of this study relates to the use of three different recruitment strategies. This was a pragmatic choice to facilitate achievement of a sufficient number of respondents in order to power the statistical analysis [25]. However, a sensitivity analysis did not identify any systematic differences in respondents' preferences for the attribute levels depending on recruitment source.

Another limitation of this study may be the statistically significant differences in gender and time to onset of drug effect in the patient sample (i.e. receiving the educational tool or the plain text). Therefore, these characteristics were tested as interactions in the RPL model that did not statistically influence the respondents' preferences for the attribute levels.

Patients responding to this study were probably relatively experienced with regard to the disease and different treatment alternatives and therefore relatively knowledgeable for patients participating in a DCE (i.e. half of the respondents had their RA for more than 10 years). In this regard, informing respondents using an educational tool might be more efficient when the decision context is not previously well known to the respondents.

4.2 Conclusions

A key concern in including patient perspectives in medical decision-making is to make sure that the patients have understood what is at stake so they can make a more informed choice.

Respondents informed by the educational tool placed relatively more importance on treatment side effects, and respondents receiving the plain text placed relatively more importance on the treatment effectiveness and the administration methods. There were no differences in reported difficulty or the time taken to answer the DCE questions between the study arms. There is still uncertainty regarding how patient preferences, and the results of a DCE study are affected by educational tools. Further research is needed to provide guidance on the use of educational tools in preference elicitation studies in respondents with less previous knowledge.

4.3 Practice implications

At this point, there is still uncertainty regarding how educational tools influence patients' preferences for treatment alternatives. Clearly, further research needs to explain the mechanisms by which educational tools influence patients' treatment preferences. Furthermore, future research is needed to determine why graphics, pictograms, icon arrays, spoken text, and click-on functions appear to change patients' preferences. Such additional research will provide guidance on how and when to introduce educational tools in assessments of patient preferences.

Ethical approval

The study was approved by the regional ethics review board in Uppsala, Sweden (Reg no. 2017/521, 2018/156). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study. I confirm all patient/personal identifiers have been removed or disguised so the patient/person(s) described are not identifiable and cannot be identified through the details of the story.

Conflict of interest

KSB, JV, MH, EB, MF and UK have no conflict of interest to declare. KR is supported by the Birmingham NIHR Biomedical Research Centre and is a member of the Research into Inflammatory Arthritis Centre Versus Arthritis and the MRC Versus Arthritis Centre for Musculoskeletal Ageing Research.

References

1. de Bekker-Grob EW, Berlin C, Levitan B, Raza K, Christoforidi K, Cleemput I et al. Giving Patients' Preferences a Voice in Medical Treatment Life Cycle: The PREFER Public-Private Project. Patient. 2017;10(3):263-6. doi:10.1007/s40271-017-0222-3.

2. Janssens R, Russo S, van Overbeeke E, Whichello C, Harding S, Kubler J et al. Patient Preferences in the Medical Product Life Cycle: What do Stakeholders Think? Semi-Structured Qualitative Interviews in Europe and the USA. Patient. 2019;12(5):513-26. doi:10.1007/s40271-019-00367-w.

3. Viney R, Savage E, Louviere J. Empirical investigation of experimental design properties of discrete choice experiments in health care. Health Econ. 2005;14(4):349-62. doi:10.1002/hec.981.

4. Ryan M, Gerard K. Using discrete choice experiments to value health care programmes: current practice and future research reflections. Appl Health Econ Health Policy. 2003;2(1):55-64.

5. Harrison M, Rigby D, Vass C, Flynn T, Louviere J, Payne K. Risk as an attribute in discrete choice experiments: a systematic review of the literature. Patient. 2014;7(2):151-70. doi:10.1007/s40271-014-0048-1.

6. Vass C, Rigby D, Payne K. "I Was Trying to Do the Maths": Exploring the Impact of Risk Communication in Discrete Choice Experiments. Patient. 2019;12(1):113-23. doi:10.1007/s40271-018-0326-4.

7. Vass CM, Davison NJ, Vander Stichele G, Payne K. A Picture is Worth a Thousand Words: The Role of Survey Training Materials in Stated-Preference Studies. Patient. 2020;13(2):163-73. doi:10.1007/s40271-019-00391-w.

8. Fraenkel L, Nowell WB, Michel G, Wiedmeyer C. Preference phenotypes to facilitate shared decision-making in rheumatoid arthritis. Ann Rheum Dis;77(5):678-83. 2018. doi:10.1136/annrheumdis-2017-212407.

9. Ho MP, Gonzalez JM, Lerner HP, Neuland CY, Whang JM, McMurry-Heath M et al. Incorporating patient-preference evidence into regulatory decision making. Surg Endosc. 2015;29(10):2984-93. doi:10.1007/s00464-014-4044-2.

10. Stacey D, Legare F, Lewis K, Barry MJ, Bennett CL, Eden KB et al. Decision aids for people facing health treatment or screening decisions. Cochrane Database Sys Revie. 2017;4:Cd001431. doi:10.1002/14651858.CD001431.pub5.

11. Meara A, Crossnohere NL, Bridges JFP. Methods for measuring patient preferences: an update and future directions. Curr Opin Rheumatol. 2019;31(2):125-31. doi:10.1097/bor.0000000000000587.

12. Ho MP, Gonzalez JM, Lerner HP, Neuland CY, Whang JM, McMurry-Heath M et al. Incorporating patient-preference evidence into regulatory decision making. Surg Endosc. 2015;29(10):2984-93.

13. Verschueren S, Buffel C, Vander Stichele G. Developing Theory-Driven, Evidence-Based Serious Games for Health: Framework Based on Research Community Insights. JMIR serious games. 2019;7(2):e11565. doi:10.2196/11565.

14. Ozdemir S. Improving the Validity of Stated-Preference Data in Health Research: The Potential of the Time-to-Think Approach. Patient. 2015;8(3):247-55. doi:10.1007/s40271-014-0084-x.

15. Lancsar E, Louviere J, Flynn T. Several methods to investigate relative attribute impact in stated preference experiments. Soc Sci Med (1982). 2007;64(8):1738-53. doi:10.1016/j.socscimed.2006.12.007.

16. Bridges JF, Hauber AB, Marshall D, Lloyd A, Prosser LA, Regier DA et al. Conjoint analysis applications in health-a checklist: a report of the ISPOR Good Research Practices for Conjoint Analysis Task Force. Value Health. 2011;14(4):403-13. doi:10.1016/j.jval.2010.11.013.

17. Wright SJ, Vass CM, Sim G, Burton M, Fiebig DG, Payne K. Accounting for Scale Heterogeneity in Healthcare-Related Discrete Choice Experiments when Comparing Stated Preferences: A Systematic Review. Patient. 2018;11(5):475-88. doi:10.1007/s40271-018-0304-x.

18. Wangdahl JM, Martensson LI. The communicative and critical health literacy scale--Swedish version. Scand J of Public Health. 2014;42(1):25-31. doi:10.1177/1403494813500592.

19. McNaughton CD, Cavanaugh KL, Kripalani S, Rothman RL, Wallston KA. Validation of a Short, 3-Item Version of the Subjective Numeracy Scale. Med Decis Making. 2015;35(8):932-6. doi:10.1177/0272989x15581800.

20. Swait J, & Louviere, J. The Role of the Scale Parameter in the Estimation and Comparison of Multinomial Logit Models. J Mark Res. 1993;30(3):305-14. doi:10.2307/3172883.

21. Housten AJ, Kamath GR, Bevers TB, Cantor SB, Dixon N, Hite A et al. Does Animation Improve Comprehension of Risk Information in Patients with Low Health Literacy? A Randomized Trial. Med Decis Making. 2020;40(1):17-28. doi:10.1177/0272989x19890296.

22. Veldwijk J, Groothuis-Oudshoorn CGM, Kihlbom U, Langenskiöld S, Dekker E, Kallenberg FGJ et al. How psychological distance of a study sample in discrete choice experiments affects preference measurement: a colorectal cancer screening case study. Patient Prefer Adherence. 2019;13:273-82. doi:10.2147/ppa.S180994.

23. Hamilton RW TD. Is there a substitute for direct experience? Comparing consumers' preferences after direct and indirect product experiences. J. Consum. Res. 2007;34(4):546–55. doi:https://doi.org/10.1086/520073.

24. Trope Y, Liberman N. Temporal construal and time-dependent changes in preference. J Pers and Soc Psychol. 2000;79(6):876-89. doi:10.1037//0022-3514.79.6.876.

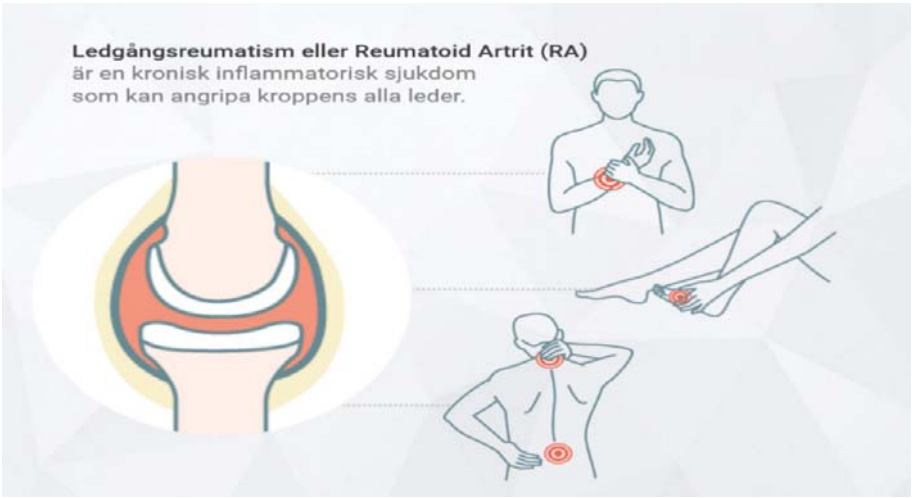
25. de Bekker-Grob EW, Donkers B, Jonker MF, Stolk EA. Sample Size Requirements for Discrete-Choice Experiments in Healthcare: a Practical Guide. Patient. 2015;8(5):373-84. doi:10.1007/s40271-015-0118-z.

Appendix: Supplementary material

Training material in plain text. Some of the illustrations from the educational tool are added to provide insight in how the educational tool were designed.

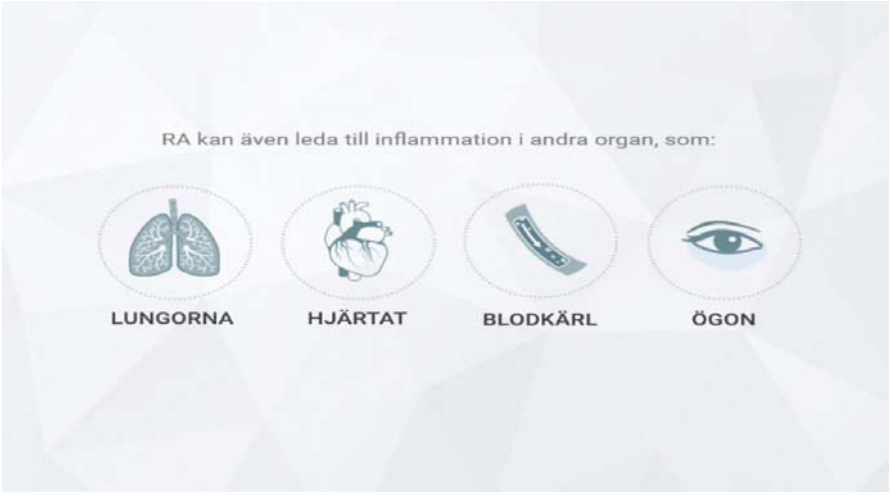
Rheumatoid arthritis

Rheumatoid arthritis (RA) as it is also called a chronic inflammatory disease that can attack all joints in the body.



Damage to the joints can occur early during the course of the disease.

If uncontrolled, this can lead to deformity of the joints or decrease in mobility. RA can also lead to inflammation in other organs, such as the lungs, the heart, blood vessels, and eyes.



Treatment

Currently, there is no cure for RA, but there are treatments. The treatment goal is to control the inflammation in the joints as much as possible and prevent joint damage. Treatment also reduces stiffness, pain, and fatigue and improves mobility and muscle strength.

In the long term, other negative effects of the disease can be reduced or prevented, such as osteoporosis and vein degradation. A successful treatment allows RA patients to keep working and live life as usual.

Treatment strategies

Several factors affect RA. Therefore, a combination of different drugs inhibit the disease in

different ways. Different drugs work for different people. The earlier treatment is started, the greater the ability to control the disease and reduce the risk of damage.

What treatment suits you?

Treatment that controls inflammation is called anti-rheumatic drugs: synthetics, biologics, and targeted synthetics. They are all characterised in different ways such as how they are taken, their side effects, and their effects. Patience may be needed to find the most effective treatment. You may also experience side effects that make you need to stop taking a medicine.

Your treatment option

We are interested in knowing what treatment you would choose. This is why ewe asdk you to choose between treatment options in part 2.

Imagine the following situation:

Your current treatment is not working as you want it to, you might get the full effect or you are experiencing unbearable side effects that you cannot stand.

You now need to change treatment:

On the next page, you can select different characteristics to find out more about them such as more information about how anti-rheumatic drugs work and the choices you will face.

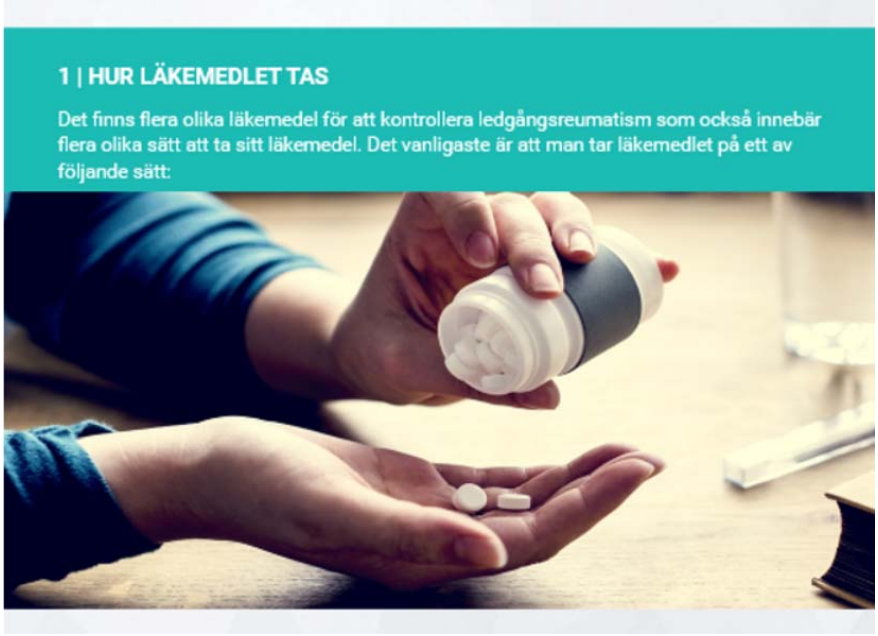
In the following part (part 2 choice tasks), we present 15 treatment choices to you. Those choices consist of 15 hypothetical RA treatment alternatives. Please indicate for every choice question the treatment alternative you would prefer.

Attributes and attribute levels

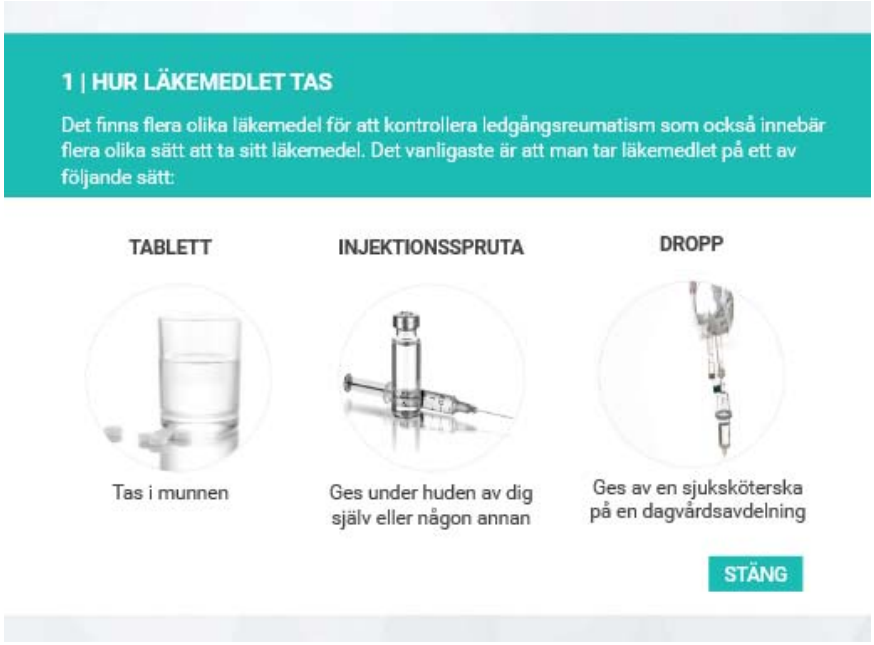

EGENSKAPER hos antireumatiska läkemedel.
Klicka på siffrorna.
Hur läkemedlet tas
1
2
Hur ofta läkemedlet tas
3
Risk för mildare övergående biverkningar
4
Risk för yttre biverkningar
5
Risk för psykiska biverkningar
6
Risk för allvarligare biverkningar
7
Effektivitet

How to take the medicine

Different treatments for RA involve different ways of taking the medicine. The most common methods are as follows:

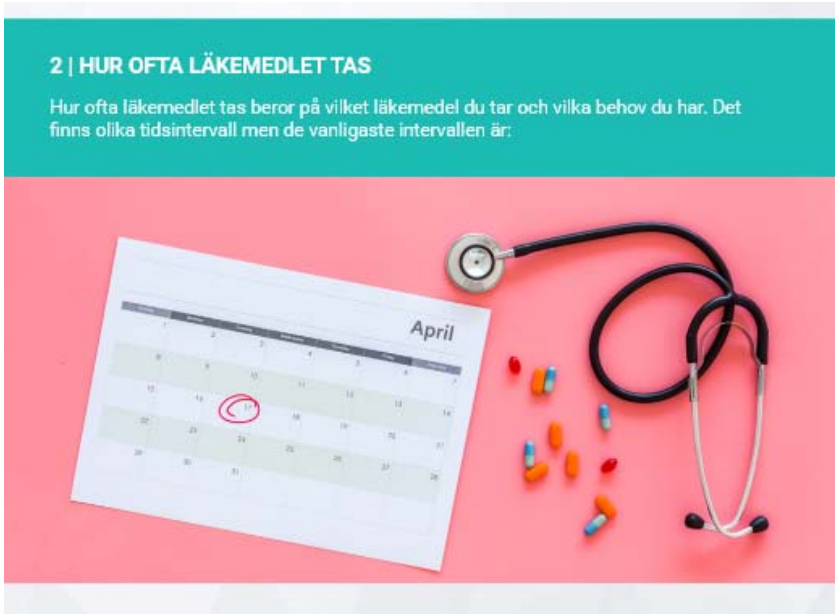


- Tablet: taken orally
- Injection: injected by you or by someone else
- Infusion (or drip): given by a nurse in a day care department

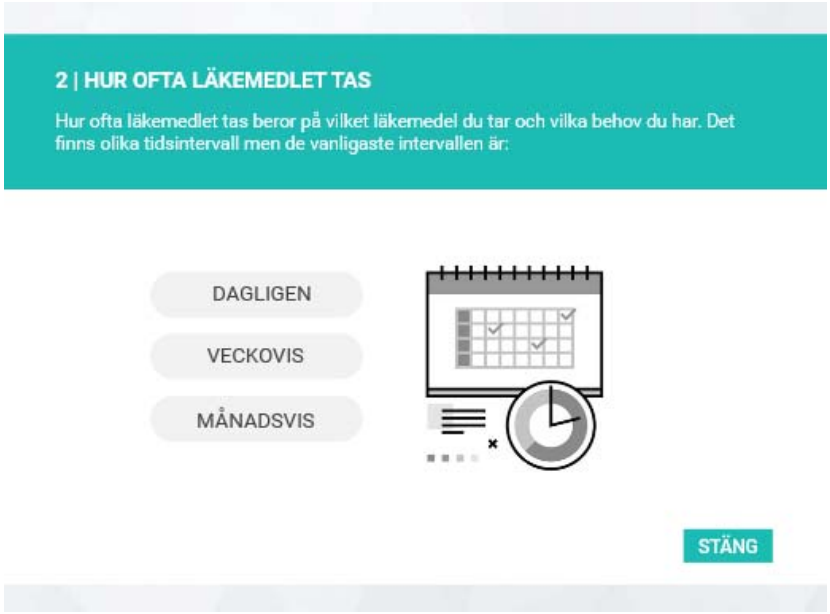


How often to take the medicine

Some medicines need to be taken more frequently than others. The most common frequencies are as follows:



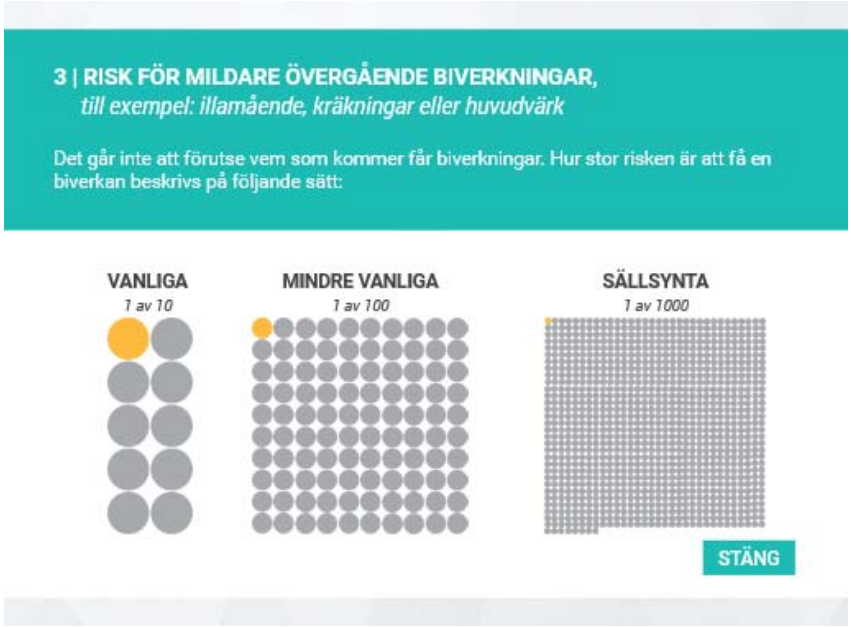
- Daily
- Weekly
- Monthly



Risk of mild short-term side effects such as nausea, vomiting, or headache

Medicines may have short-term side effects that pass away after a while and are not life threatening. These side effects can include nausea, vomiting, and headaches. It is not possible to predict who will experience these side effect. The risk of side effects can be described as follows:

- Common: 1 out of 10: so out of every 10 patients who take the drug, 1 suffers from the side effect
- Uncommon: 1 out of 100: so out of every 100 patients who take the drug, 1 suffers from the side effect
- Rare: 1 out of 1000: so out of every 1000 patients who take the drug, 1 suffers from the side effect



Risk of side effects affecting appearance such as hair loss, weight gain, or skin rash

Some medicines may have side effects that change your appearance. These side effects include hair loss, weight gain, and skin rashes. It is not possible to predict who will experience these side effects. The risk of side effects affecting your appearance can be described as follows:

- Common: 1 out of 10: so out of every 10 patients who take the drug, 1 suffers from the side effect
- Uncommon: 1 out of 100: so out of every 100 patients who take the drug, 1 suffers from the side effect
- Rare: 1 out of 1000: so out of every 1000 patients who take the drug, 1 suffers from the side effect

Risk of psychological side effects such as anxiety, mood changes, depression, and sleep disturbance

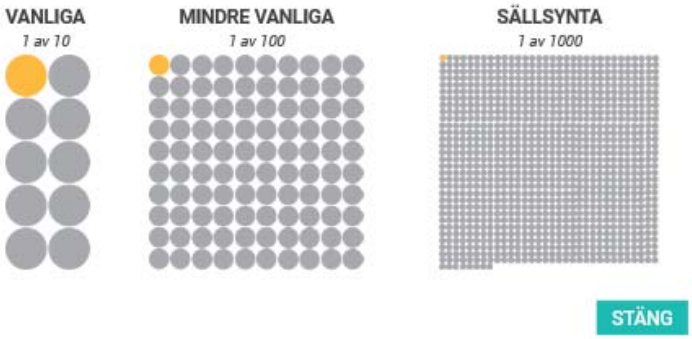
Some medicines can lead to anxiety, depression, or sleep disturbances. It is not possible to predict who will experience these side effects. The risk of these kinds of side effect can be described as follows:

- Common: 1 out of 10: so out of every 10 patients who take the drug, 1 suffers from the side effect
- Uncommon: 1 out of 100: so out of every 100 patients who take the drug, 1 suffers from the side effect
- Rare: 1 out of 1000: so out of every 1000 patients who take the drug, 1 suffers from the side effect

5 | RISK FÖR PSYKISKA BIVERKNINGAR

till exempel: nedstämdhet, humörförändringar, depression eller sömnsvårigheter

Det går inte att förutse vem som kommer får biverkningar. Hur stor risken är att få en biverkan beskrivs på följande sätt:

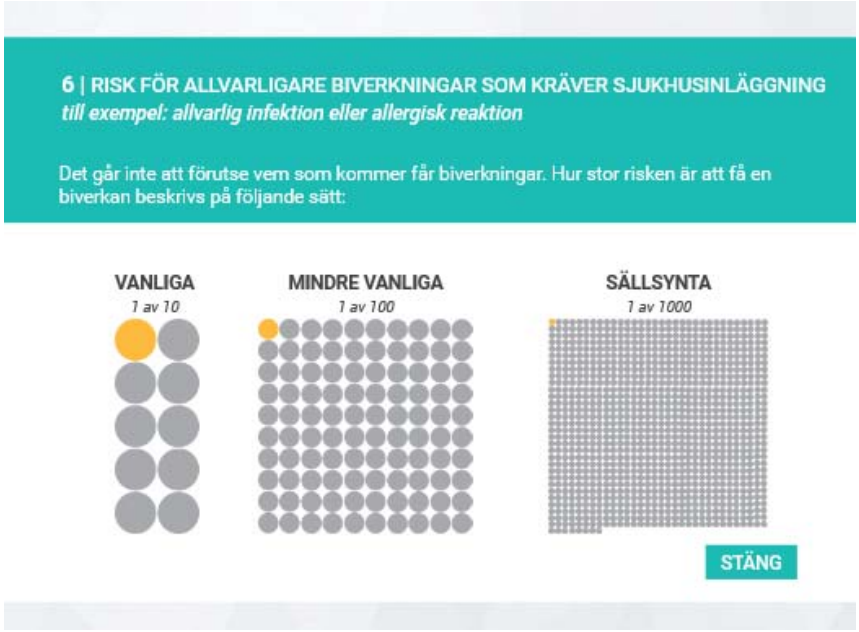


STÄNG

Risk of more severe side effects that require hospitalization such as severe infections or allergic reactions

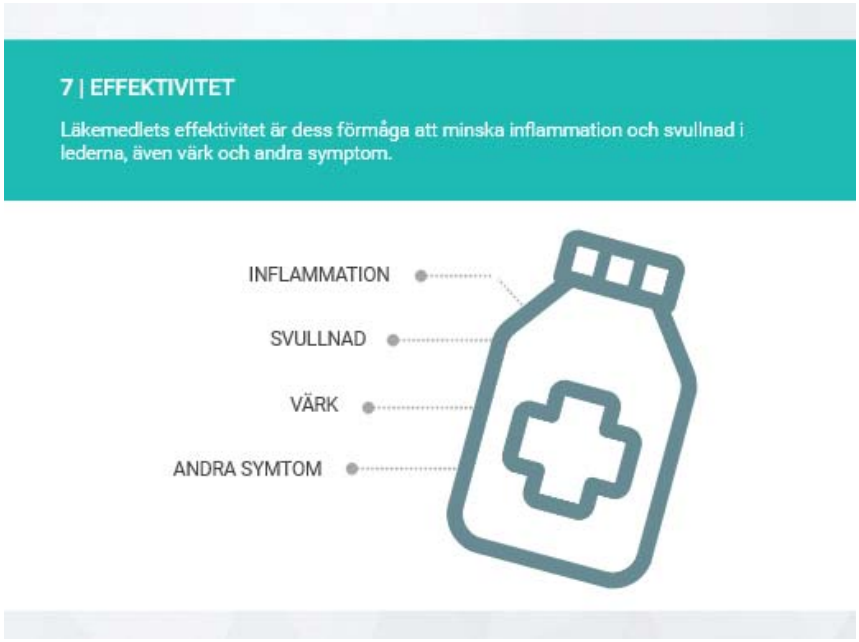
Some medicines can have serious side effects that may be life threatening and may require you to be treated in a hospital. These side effects can include serious infections, allergic reactions, and sepsis. It is not possible to predict who will experience these side effects. The risk of these side effects can be described as follows:

- Common: 1 out of 10: so out of every 10 patients who take the drug, 1 suffers from the side effect
- Uncommon: 1 out of 100: so out of every 100 patients who take the drug, 1 suffers from the side effect
- Rare: 1 out of 1000: so out of every 1000 patients who take the drug, 1 suffers from the side effect



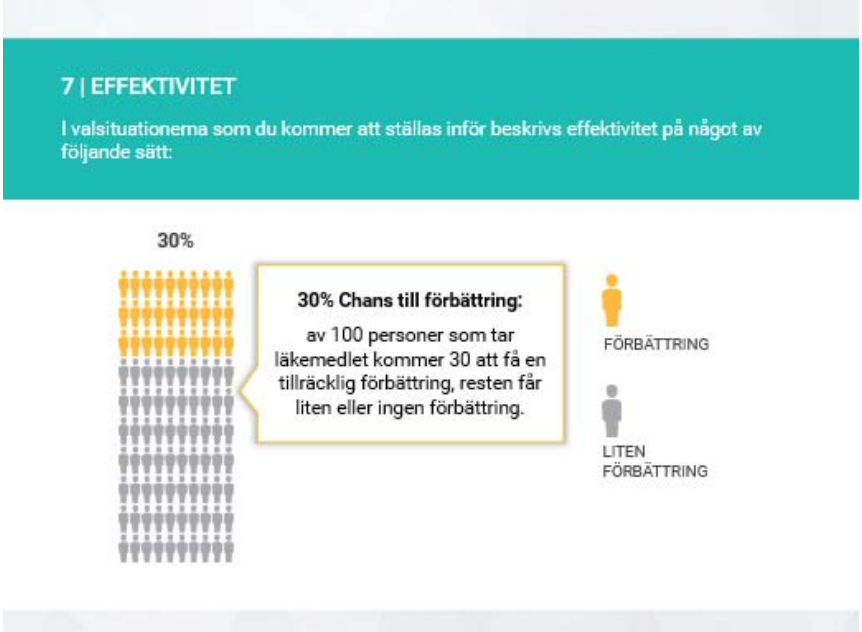
Effectiveness

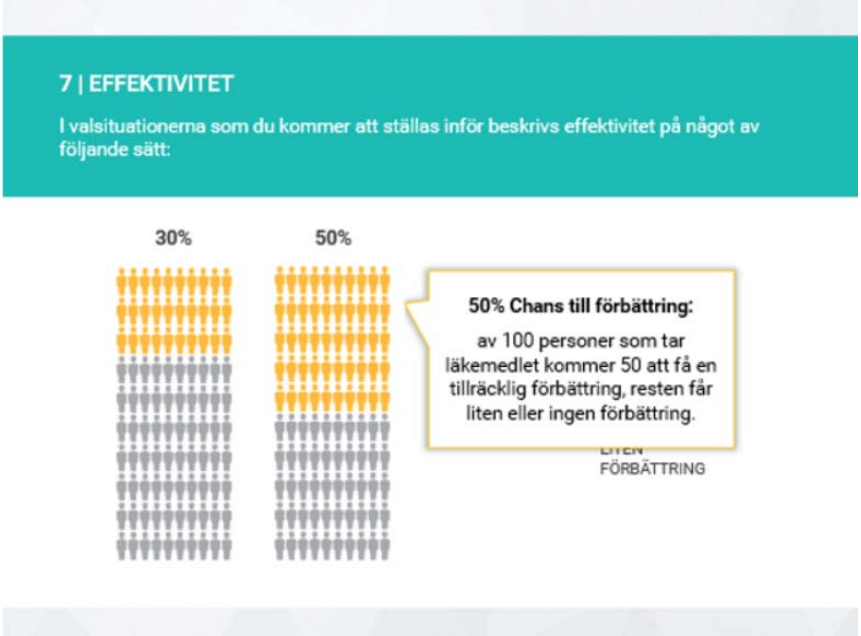
The effectiveness of the treatment is the ability to control inflammation and swelling in the joints and pain and other symptoms. In the choice situations, the effectiveness of the treatment will be described as follows:

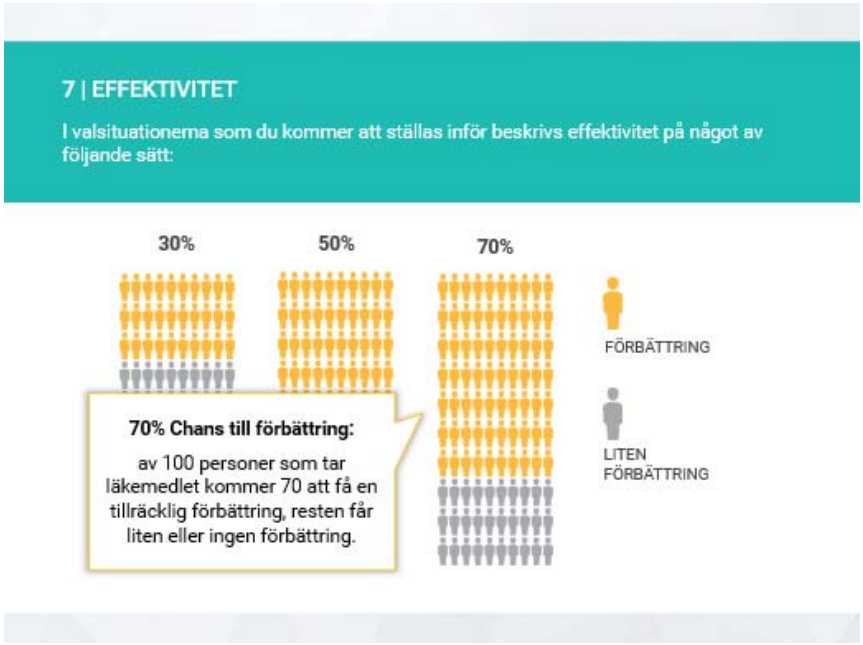


- 30% Chance of improvement: so out of 100 persons taking the treatment, 30 will experience enough improvement, the rest will experience a small or no improvement.

- 50% Chance of improvement: so out of 100 persons taking the treatment, 50 will experience enough improvement, the rest will experience small or no improvement.
- 70% Chance of improvement: so out of 100 persons taking the treatment, 70 will experience enough improvement, the rest will experience a small or no improvement.







By ‘enough improvement’ we mean 50% improvement from starting point when the treatment is not working. A ‘small improvement’ is <20% from the starting point.