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## Psychometric characteristics of the Revised Illness Perception Questionnaire (IPQ-R) in adults with sickle cell disease

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### ABSTRACT

**Objective:** Sickle cell disease (SCD) is the most frequent monogenic disease worldwide. Psychological and behavioural factors are often reported as playing a significant role in predicting SCD health outcomes. When focusing on adaptation to a specific health condition and its treatment, the Common Sense Model of Health and Illness (CSM) has proven to be of heuristic value. In other health conditions, illness outcomes are directly influenced by illness perception. Therefore, the aim of this study is to explore the psychometric proprieties of the Revised Illness Perception Questionnaire (IPQ-R).

**Design:** We performed a cross-sectional assessment on 517 adult patients with sickle cell disease and collected the results of 406 IPQ-R. With these data, we verified the factor structure of the Belief scale and proposed modifications to improve its fit to the data with a confirmatory factor analysis. In addition, we explored the factorial structure of the Causal attribution scale with an exploratory factor analysis.

**Results:** The initial model showed poor fit with the data. After structural modifications, elimination of two items with a low loading (model 2), covariance added between items (model 3) and items reallocation (model 4), the last model proposed presented a correct fit with the data. Before doing this model specification, we reviewed and compiled the nine studies that explored the psychometric properties of the IPQ-R in order to highlight all the modifications made by the other authors who have adapted the IPQ-R to a specific population and to allow a comparison with our own modifications.

**Conclusion:** Considering previous findings, this research suggests further work is needed on the structure of the dimensions of the IPQ-R.

### ARTICLE HISTORY

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### KEYWORDS

Sickle cell disease; Illness Perception Questionnaire-Revised; confirmatory factor analysis; psychometric; Common Sens Model

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## 1. Introduction

### 1.1 Background

Sickle cell disease (SCD) is the most frequent monogenetic disease worldwide (Piel, Steinberg, & Rees, 2017), characterised by chronic haemolytic anaemia, painful episodes of vaso-occlusive crises (VOC), progressive organ failure and reduced life expectancy (Houwing et al., 2019). Therapeutic options for patients remain scarce although haematopoietic stem cell transplantation and gene therapy are receiving greater attention (Tanhehco & Bhatia, 2019). In high-income countries, comprehensive care programmes have increased the life expectancy of SCD patients, with more than 99% of patients now reaching adulthood (Gardner et al., 2016). Creating a model of lifespan care, which would allow a transition from an acute disease to a chronic condition through preventive measures, is getting more and more attention for determining health outcomes (Minniti & Vichinsky, 2017).

The health outcomes experienced by adults with SCD are largely determined by their ability to carefully manage their condition (Levenson, 2008), including their ability to adhere to treatment recommendations (Loiselle et al., 2016). It is believed that up to 75% of patients struggle with adherence to treatment regimens (Oudin-Dogliani, Gay, Lehougre, Arlet, & Galactéros, 2019). For instance, hydroxyurea (HU) has proved to be of high efficacy in reducing symptomatic manifestations of SCD (Voskaridou et al., 2010). Nevertheless, its use and adherence remain suboptimal (Lanzkron, Haywood, Segal, & Dover, 2006; Okam, Shaykevich, Ebert, Zaslavsky, & Ayanian, 2014; Shankar et al., 2005).

Health outcomes are predicted by the Common Sense Model of Health and Illness (CSM), a schema that has proved useful in understanding how patients' perception of their illness guide coping behaviour including diseases self-management and adherence to treatment (Hagger, Koch, Chatzisarantis, & Orbell, 2017; Hagger & Orbell, 2003; Leventhal et al., 1997). For example, in the case of therapeutic adherence, individuals living with SCD may attempt to assign meanings about SCD, i.e. expectation about its cause, or its future controllability, beliefs about its negative consequences and evolution over time. In turn, these have repercussions on the coping strategies, which could be linked to a poorer adherence.

In other health conditions, illness outcomes are directly influenced by illness perception (Hagger & Orbell, 2003), although the pattern of association between illness perceptions and health outcome is a sample and/or illness-dependent.

Growing evidence from different illnesses indicates that intervention can be effective in modifying maladaptive illness perception, thus improving patients' health-related behaviour and outcomes (Broadbent, Ellis, Thomas, Gamble, & Petrie, 2009; Petrie, Perry, Broadbent, & Weinman, 2012; Traeger et al., 2011). So far, there is a paucity of studies investigating illness perception in people with SCD, despite a growing attention on the influence of these patients' expectations and goals on their health outcomes (Oudin-Dogliani et al., [Under editorial process](#)).

### 1.2 Aim of this study

In SCD, despite improvement in the care of the patients, adherence remain suboptimal leading to increase hospitalisations and premature death (Payne et al., 2020). The representation that patients have of their own disease or illness could explain or predict

health behaviour, specifically adherence (Kucukarslan, 2012). In order to implement theory-driven interventions oriented to adults with SCD we need to adapt the IPQ-R to our population to correctly evaluate its perception of SCD. Despite previous work done on adolescents with SCD (Asnani, Barton-Gooden, Grindley, & Knight-Madden, 2016), to our knowledge, Illness Perception Questionnaire (IPQ-R) has not yet been adapted to adults with SCD. The objective of this research was to adapt the IPQ-R to be relevant for use in adults with SCD and to evaluate the factor structure of the modified instrument.

## 2. Materials and method

### 2.1 Participants

Patients were recruited from the French referral centre for sickle cell disease at Henri Mondor hospital (UMGGR). Eligibility criteria were homozygous or heterozygous SCD adult patients. Recruitment was undertaken by a clinical psychologist and a research assistant nurse, who identified and approached eligible patients, distributed study information and obtained written informed consent. Patients were approached in person, typically during outpatient appointments.

### 2.2 Evaluation of illness perceptions: Illness Perception Questionnaire (IPQ)

Illness perceptions as postulated in the CSM are usually measured with different forms of the Illness Perception Questionnaire (IPQ) (Weinman, Petrie, Moss-Morris, & Horne, 1996) or its later form obtained after factorial extension from five to nine dimensions: IPQ-Revised (IPQ-R) (Moss-Morris et al., 2002). A shorter form of the revised version is also available: B-IPQ (Broadbent, Petrie, Main, & Weinman, 2006). The most frequently used form is the revised version IPQ-R. IPQ-R is composed of three separate sections.

#### 2.2.1 Description of the three sections

The separation into three sections appeared with the revision of the IPQ by Moss-Morris et al. (2002). This separation does not correspond to the existence of a hierarchical structure which would include three over-arching domains each composed of several sub-domains. Scores are provided for domains only and there are no total scores.

**2.2.1.1 The Identity scale.** The *Identity scale* presents a list of the 12 commonly experienced symptoms included in the original version of IPQ (e.g. pain, nausea, sleep difficulty) plus two new symptoms (sore throat and wheeziness). The Identity scale has a dichotomous rating with a yes/no system. In the first part, patients must rate whether they have experienced one of the 14 symptoms enlisted and are then asked whether they believe the symptoms to be caused by or related to their illness. The sum of the yes-rated items on this second part forms the Identity subscale score. Following previous research on the psychometric validity of IPQ-R (Abubakari et al., 2012; Ashley et al., 2013; Dempster & McCorry, 2012; Hagger & Orbell, 2005; Nicholls, Hill, & Foster, 2013), we considered the Identity scale as a single factor structure. Therefore, this scale was not part of the construct validation.

**2.2.1.2 The Causal attribution scale.** The *Causal attribution scale* assesses on a Likert scale (5-point rating) the degree to which patients agree that a cause is triggering or maintaining their illness. In the revised version, four factors are described with various internal consistency: (1) psychological attribution ( $\alpha = .86$ ) that include six items (e.g. Stress or worry, overwork, my personality); (2) risk factors ( $\alpha = .77$ ) that regroup seven items (e.g. hereditary, ageing, smoking); (3) immunity ( $\alpha = .67$ ) with three items (a germ or virus, pollution in the environment, altered immunity); (4) accident or chance ( $\alpha = .23$ ) composed of two items (chance or bad luck, accident or injury) (Moss-Morris et al., 2002).

Most of the studies aiming at confirming the IPQ-R validity exclude the Causal attribution scale of the analysis. When included, mixed results are obtained: the data show a variation in the number of factors found in this scale, ranging from 3 to 5 factors depending on the chronic disease (Abubakari et al., 2012; Hagger & Orbell, 2005; Nicholls et al., 2013). Generally, causes that could be perceived as triggering or maintaining an illness are condition-specific meaning that this scale should be adapted to the specific disease and that a new exploratory appraisal of the factorial structure is needed.

**2.2.1.3 The beliefs scale.** The *Beliefs scale* is composed of 38 items rated in a five-point Likert scale and regrouped in seven core factors, displayed below. Overall internal consistency in the original factorial analysis by Moss-Morris et al. (2002) is good.

- Timeline acute/chronic (chronology) ( $\alpha = .89$ ) composed of six items (m/M [6; 30]), investigates the beliefs about the duration of the illness.
- Timeline cyclical (Cyclical) ( $\alpha = .79$ ), composed of four items (m/m [4; 20]), explores whether the illness trajectory is constant or cyclical.
- Consequences ( $\alpha = .84$ ), composed of six items (m/M [6; 30]), assess the impact of the illness on the patient's life.
- Personal control (controllability) ( $\alpha = .81$ ) composed of six items (m/M [6; 30]), evaluate the perceived influence patient thought to have on the illness.
- Treatment control (cure) ( $\alpha = .80$ ), composed of five items (m/M [5; 25]), estimates the perceived treatment efficacy.
- Illness coherence (coherence) ( $\alpha = .87$ ), composed of five items (m/M [5; 25]), investigate how well patients understand their illness.
- Emotional representation ( $\alpha = .88$ ), composed of six items (m/M [6; 30]), explores the emotional impact of the illness.

The interpretation that can be made of a high score in a dimension is summarised below (Table 1).

## 2.2.2 Previous psychometric validation and modifications

The IPQ-R has been used in different clinical setting with acute or chronic conditions but few studies have validated its psychometric proprieties (Abubakari et al., 2012; Ashley et al., 2013; Chateaux & Spitz, 2006; Dempster & McCorry, 2012; Ferreira, Gay, Regnier-Aeberhard, & Bricaire, 2010; Hagger & Orbell, 2005; Horne & Weinman, 2002; Koehler, Koenigsmann, & Frommer, 2009; Nicholls et al., 2013; Sachsa, 2010; Surgenor et al., 2019; Taylor, O'Neill, Hughes, & Moss-Morris, 2018).

**Table 1.** Interpretation of IPQ-R dimension scores (from Nicholls et al., 2013).

Dimensions	Interpretation
<i>A high dimension indicates stronger agreement that ...</i>	
Timeline acute/chronic	Illness will last a long time
Timeline cyclical	Condition is cyclical in nature
Consequences	Condition has a major impact on life
Personal control	Good perceived personal control over symptoms
Treatment control/Cure	Treatment could control symptoms
Illness coherence	Understanding of condition
Emotional representation	Condition affects patients emotionally

From 2005 to 2019, nine studies have inspected the psychometric proprieties of the IPQ-R (Abubakari et al., 2012; Ashley et al., 2013; Brzoska, Yilmaz-Aslan, Sultanoglu, Sultanoglu, & Razum, 2012; Cabassa, Lagomasino, Dwight-Johnson, Hansen, & Xie, 2008; Dempster & McCorry, 2012; Hagger & Orbell, 2005; Nicholls et al., 2013; Surgenor et al., 2019; Wittkowski, Richards, Williams, & Main, 2008). Although it allowed identification of the seven core dimensions, multiple modifications were made to fulfil confirmatory factor analysis requirements (Table 2). However, since the seven dimensions appear constant in different populations, IPQ-R could be successfully adapted to other specific conditions. Following the suggestions of the IPQ-R developers, adaptation typically includes wording changes to substitute the specific context.

Of the nine studies that have evaluated the original factor structure of the IPQ-R, only one did not specify modifications to the initial model (Wittkowski et al., 2008). For the others, a combination of three modifications were performed: deletion of items with factor loading under 0.40, specifying errors covariances, and reallocation of items to their original dimension to another dimension. With the exception of Wittkowski *et al.*, all the final models had satisfactory fitting indices: the comparative fit index (CFI) range was comprised within [.859; 0.95], the Tucker-Lewis index (TLI) (NNFI) [0.90; 0.921], the standardised root mean squared residual SRMSR within [.06; .079], the root mean square of approximation (RMSEA) within [.037; .065], and the relative  $\chi^2$  ( $\chi^2$ /df) (Wheaton, Muthen, Alwin, & Summers, 1977) within [1.435; 7.801].

### 2.2.3 Adaptation to SCD

Data were collected using the three sections. We used the IPQ-R with wording modification (replacement of the generic term ‘disease’ by ‘sickle cell disease’) to adapt to SCD in the Beliefs scale. The Identity scale was not modified. The Causal attribution scale was extended to reflect the specific beliefs adults with SCD could have following previous work by Gernet, Mestre, and Runel-Belliard (2011) and Oudin-Dogliani et al. (2019). A medical validation of the questionnaire was carried out, in the same way, the comprehensibility and the feasibility of the questionnaire were verified with two patients leading therapeutic patient education programme.

We followed a careful cross-cultural translation and validation methodology (Cha, Kim, & Erlen, 2007). Discrepancies emerging from the back translation were discussed, and adjustments to the translation were made (Table 3).

In addition to completing the IPQ-R, patients provided demographic information (age, gender, ethnicity) and medical information (genotype).

**Table 2.** Comparison of the goodness of fit indices between the nine studies that explored the psychometric properties of the IPQ-R.

Initial model	n	Indices of goodness of fit for the original CFA					AIC	Comment
		CFI	TLI	SRMSR	RMSEA	$\chi^2/df$		
Surgenor, L. J., Snell, D. L., Siegert, R. J., Kelly, S., Flint, R., & Coulter, G. (2019).	310	> 0.90 ?	> 0.90 ?	<0.08 ?	<0.07 ?	<5 ?	?	No data provided
Ashley, L., Smith, A. B., Keding, A., Jones, H., Velikova, G., & Wright, P. (2013).	531	0.80		0.086	0.042	1.766		
Nicholls, E. E., Hill, S., & Foster, N. E. (2013).	330 1621 1319	0.86 0.86 0.90			0.064 0.068 0.059	2.338 8.495 5.635		
Brzoska, P., Yilmaz-Aslan, Y., Sultanoglu, E., Sultanoglu, B., & Razum, O. (2012).	302	0.785	0.765	0.109	0.075	3.722	18177.43	
Abubakari, A.-R., Jones, M. C., Lauder, W., Kirk, A., Devendra, D., & Anderson, J. (2012).	221	0.83	0.81		0.06	1.707		
Dempster, M., & McCorry, N. K. (2012).	587	0.94		0.070	0.068	3.707		
Cabassa, L. J., Lagomasino, I. T., Dwight-Johnson, M., Hansen, M. C., & Xie, B. (2008).	339	0.793	0.768		0.069	2.598		
Wittkowski, A., Richards, H. L., Williams, J., & Main, C. J. (2008).	284	0.798			0.085	3.061		
Hagger, M. S., & Orbell, S. (2005).	660	0.911	0.904		0.042			
<b>Modification 1 = items deletion</b>	<b>n</b>		<b>Indices of goodness of fit for the original CFA</b>				<b>AIC</b>	<b>Items deleted</b>
		<b>CFI</b>	<b>TLI</b>	<b>SRMSR</b>	<b>RMSEA</b>	<b><math>\chi^2/df</math></b>		
		> 0.90 ?	> 0.90 ?	<0.08 ?	<0.07 ?	<5 ?		
Surgenor, L. J., Snell, D. L., Siegert, R. J., Kelly, S., Flint, R., & Coulter, G. (2019).	310							19, 23
Ashley, L., Smith, A. B., Keding, A., Jones, H., Velikova, G., & Wright, P. (2013).	531	0.904		0.058	0.057	2.722		12, 18, 24, 32
Nicholls, E. E., Hill, S., & Foster, N. E. (2013).	330 1621 1319	0.89 0.90 0.92			0.064 0.065 0.058	2.344 7.801 5.452		15, 17, 19, 23, 37, 38
Brzoska, P., Yilmaz-Aslan, Y., Sultanoglu, E., Sultanoglu, B., & Razum, O. (2012).	302	0.877	0.864	0.083	0.059	2.924	15404.43	17, 19, 20, 31
	221	0.86	0.85		0.06	1.669		19, 20, 24,

			Indices of goodness of fit for the original CFA					AIC	Item with error covarianced
		CFI	TLI	SRMSR	RMSEA	$\chi^2/df$			
		> 0.90	> 0.90	<0.08	<0.07	<5			
Abubakari, A.-R., Jones, M. C., Lauder, W., Kirk, A., Devendra, D., & Anderson, J. (2012).									
Dempster, M., & McCorry, N. K. (2012).	587	0.94		0.070	0.068	3.707			
Cabassa, L. J., Lagomasino, I. T., Dwight-Johnson, M., Hansen, M. C., & Xie, B. (2008).	339	0.827	0.802		0.069	2.620		15, 17, 19	
Wittkowski, A., Richards, H. L., Williams, J., & Main, C. J. (2008).	284	0.798			0.085	3.061			
Hagger, M. S., & Orbell, S. (2005).	660	0.926	0.920		0.040			9, 10	
<b>Modification 2 = adding error covariances</b>	<b>n</b>								
		> 0.90	> 0.90	<0.08	<0.07	<5			
Surgenor, L. J., Snell, D. L., Siegert, R. J., Kelly, S., Flint, R., & Coulter, G. (2019).	310	?	?	?	?	?	?	1&4 6&7 6&9 9&11 10&11 12&17 15&17 34&37 37&38	
Ashley, L., Smith, A. B., Keding, A., Jones, H., Velikova, G., & Wright, P. (2013).	531	0.859		0.079	0.037	1.526		1&4 15&17 19&23	
Nicholls, E. E., Hill, S., & Foster, N. E. (2013).	330 1621 1319	0.89 0.90 0.92			0.064 0.065 0.058	2.344 7.801 5.452			
Brzoska, P., Yilmaz-Aslan, Y., Sultanoglu, E., Sultanoglu, B., & Razum, O. (2012).	302	0.911	0.901	0.079	0.050	2.502	15189.58	33&34 7&8	
Abubakari, A.-R., Jones, M. C., Lauder, W., Kirk, A., Devendra, D., & Anderson, J. (2012).	221	0.89	0.88		0.05	1.522		1&4 7&6 6&8 7&8 9&11 34&35	
Dempster, M., & McCorry, N. K. (2012).	587	0.95		0.061	0.064	3.430		1&4	
Cabassa, L. J., Lagomasino, I. T., Dwight-Johnson, M., Hansen, M. C., & Xie, B. (2008).	339	0.908	0.900		0.051	1.885		7 errors covariances non-specified	

	n	Indices of goodness of fit for the original CFA				X <sup>2</sup> /dF	AIC	Items reallocated
		CFI	TLI	SRMSR	RMSEA			
Wittkowski, A., Richards, H. L., Williams, J., & Main, C. J. (2008).	284	0.798			0.085	3.061		
Hagger, M. S., & Orbell, S. (2005).	660	0.926	0.920		0.040			
<b>Modification 3 = items reallocation (Final model)</b>								
		> 0.90	> 0.90	<0.08	<0.07	<5		
		?	?	?	?	?	?	
Surgenor, L. J., Snell, D. L., Siegert, R. J., Kelly, S., Flint, R., & Coulter, G. (2019).	310							
Ashley, L., Smith, A. B., Keding, A., Jones, H., Velikova, G., & Wright, P. (2013).	531	0.859		0.079	0.037	1.526		
Nicholls, E. E., Hill, S., & Foster, N. E. (2013).	330	0.89			0.064	2.344		
	1621	0.90			0.065	7.801		
	1319	0.92			0.058	5.452		
Brzoska, P., Yilmaz-Aslan, Y., Sultanoglu, E., Sultanoglu, B., & Razum, O. (2012).	302	0.929	0.921	0.067	0.045	2.282	15079.40	6 -> chronology
Abubakari, A.-R., Jones, M. C., Lauder, W., Kirk, A., Devendra, D., & Anderson, J. (2012).	221	0.91	0.90		0.05	1.435		
Dempster, M., & McCorry, N. K. (2012).	587	0.95		0.060	0.061	3.182		18 -> Treatment control
Cabassa, L. J., Lagomasino, I. T., Dwight-Johnson, M., Hansen, M. C., & Xie, B. (2008).	339	0.908	0.900		0.051	1.885		
Wittkowski, A., Richards, H. L., Williams, J., & Main, C. J. (2008).	284	0.798			0.085	3.061		
Hagger, M. S., & Orbell, S. (2005).	660	0.926	0.920		0.040			

**Table 3.** The Revised Illness Perception Questionnaire (IPQ-R) seven core-factor items and modified Causal attribution scale.

IPQ-R dimension	Item	English version	French translation
Timeline acute/ chronic	1	R My sickle cell disease will last a short time	Ma drépanocytose ne va pas durer longtemps
	2	My sickle cell disease is likely to be permanent rather than temporary	Ma drépanocytose est susceptible d'être permanente plutôt que temporaire
	3	My sickle cell disease will last for a long time	Ma drépanocytose va durer longtemps
	4	R My sickle cell disease will pass quickly	Ma drépanocytose va rapidement passer
	5	I expect to have sickle cell disease for the rest of my life	Je pense que j'aurais la drépanocytose pour le restant de ma vie
Consequences	18	R My sickle cell disease will improve in time	Ma drépanocytose va s'améliorer avec le temps
	6	My sickle cell disease is a serious condition	Ma drépanocytose est grave
	7	My sickle cell disease has major consequences in my life	Ma drépanocytose a des conséquences importantes sur ma vie
	8	R My sickle cell disease does not have much effect on my life	Ma drépanocytose n'a pas beaucoup d'effet sur ma vie
	9	My sickle cell disease strongly affects the way others see me	Ma drépanocytose affecte beaucoup la façon dont les autres me voient
	10	My sickle cell disease has serious financial consequences	Ma drépanocytose a des conséquences financières graves
Personal control	11	My sickle cell disease causes difficulties for those who are close to me	Ma drépanocytose cause des difficultés à mes proches
	12	There is a lot which I can do to control my symptoms	Il y a beaucoup de choses que je peux faire pour contrôler mes symptômes
	13	What I do can determine whether my sickle cell disease gets better or worse	Ce que je fais peut déterminer l'amélioration ou l'aggravation de ma drépanocytose
	14	The course of my sickle cell disease depends on me	Le déroulement de ma drépanocytose dépend de moi
	15	R Nothing I do will affect my sickle cell disease	Rien de ce que je fais n'affectera ma drépanocytose
	16	I have the power to influence my sickle cell disease	J'ai le pouvoir d'influencer ma drépanocytose
Treatment control	17	R My actions will have no effect on the outcome of my sickle cell disease	Mes actions n'auront aucun effet sur l'évolution de ma drépanocytose
	19	R There is very little that can be done to improve my sickle cell disease	Il y a peu de choses à faire pour améliorer ma drépanocytose
	20	My treatment will be effective in curing my sickle cell disease	Mon traitement sera efficace pour guérir ma drépanocytose
	21	The negative effects of my sickle cell disease can be prevented (avoided) by my treatment	Les effets négatifs de ma drépanocytose peuvent être évités par mon traitement
	22	My treatment can control my sickle cell disease	Mon traitement peut contrôler ma drépanocytose
Illness coherence	23	R There is nothing which can help my sickle cell disease	Rien ne peut aider mon état
	24	R The symptoms of my conditions are puzzling to me	Les symptômes de mon état me laissent perplexe
	25	R My sickle cell disease is a mystery to me	La drépanocytose est un mystère pour moi
	26	R I don't understand my sickle cell disease	Je ne comprends pas la drépanocytose
	27	R My sickle cell disease doesn't make any sense to me	La drépanocytose n'a aucun sens pour moi
Timeline cyclical	28	I have a clear picture or understanding of my condition	J'ai une image nette ou une compréhension de mon état
	29	The symptoms of my sickle cell disease change a great deal from day to day	Les symptômes de ma drépanocytose changent beaucoup d'un jour à l'autre
	30	My symptoms come and go in cycles	Mes symptômes vont et viennent par cycles
	31	My sickle cell disease is very unpredictable	Ma drépanocytose est très imprévisible

(Continued)

**Table 3.** Continued.

IPQ-R dimension	Item	English version	French translation
Emotional representation	32	I go through cycles in which my illness gets better and worse	Je passe par des cycles au cours desquels ma drépanocytose diminue ou empire
	33	I get depressed when I think about my sickle cell disease	Je déprime quand je pense à ma drépanocytose
	34	When I think about my sickle cell disease I get upset	Quand j'y pense, ma drépanocytose m'inquiète
	35	My sickle cell disease makes me feel angry	Ma drépanocytose me met en colère
	36	My sickle cell disease does not worry me	Ma drépanocytose ne me gêne pas
	37	Having this sickle cell disease makes me feel anxious	À cause de la drépanocytose, je suis anxieux
	38	My sickle cell disease makes me feel afraid	Ma drépanocytose me fait peur
Psychological attribution	44	Stress or worry	C'est le stress ou l'ennui
	45	Family problems or worries caused my illness	C'est des problèmes de familles
	55	My mental attitude e.g. thinking about life negatively	C'est mon attitude mentale ex. Avoir une vision négative de la vie.
	61	Overwork	C'est lié à ma surcharge de travail
	68	My emotional state e.g. feeling down, lonely, anxious, empty	C'est dû à mon état émotionnel ex. Ma solitude, mon anxiété ...
	70	My personality	C'est ma personnalité
Risk factor – Hereditary	39	N It runs in my family	C'est courant dans ma famille
	40	N It is a disease of the origins	C'est une maladie des origines
	51	N It is hereditary or a genetic disease	C'est héréditaire ou génétique
Risk factor – Blood	57	N It is passed from parents to children	C'est transmis des parents aux enfants
	41	N It is a lack of blood	C'est un manque de globule
	47	N It is a disease of the blood	C'est une maladie du sang
	53	N It is a poor circulation of blood	C'est une mauvaise circulation du sang
	59	N It is due to incompatible bloods	C'est dû à des sangs incompatibles
Risk factor – other	64	It is a poor quality of blood	C'est la mauvaise qualité du sang
	43	Diet or eating habits	C'est lié aux habitudes alimentaires
	46	N It is a disease from Africa	C'est une maladie de l'Afrique
	49	My own behaviour	C'est dû à mon propre comportement
	53	Poor medical care in my past	C'est dû à de mauvais soins médicaux dans mon passé personnel
	62	Ageing	C'est dû au vieillissement
Immunity	63	N It is the poor sperm quality	C'est le mauvais sperme
	65	Alcohol	C'est dû à ma consommation d'alcool
	66	Smoking	C'est dû à ma consommation de tabac
	50	A germ or virus	C'est dû à un microbe ou un virus
	52	Pollution in the environment	C'est dû à la pollution de l'environnement
	58	N It is the heat	C'est la chaleur
Accident or chance	67	Altered immunity	C'est la détérioration de l'immunité
	42	Chance or bad luck	C'est le hasard ou la malchance
	48	N It is a divine punishment	C'est une punition divine
	54	N It is a malediction	C'est une malédiction
	60	N It is due to spirits or spells e.g. Djinn, wizard, ...	C'est dû à des esprits ou à des sorts ex. Djinn, sorcier ...
	69	Accident or injury	C'est dû à un accident ou une blessure

Note: R, reverse item; N, new item.

### 2.3 Statistical analysis

Descriptive statistics summarised the demographic and medical characteristics of the sample using R.

**For the Beliefs scale**, confirmatory factor analysis (CFA) was conducted using R with package Lavaan (Rosseel & Jorgensen, 2019) under Jamovi (The Jamovi Project, 2020). We used the two-index presentation strategy (Hu & Bentler, 1999), which includes using the maximum likelihood (ML)-based standardised root mean squared residual (SRMR) with a cut-off value under .08, and supplementing it with the root mean square of approximation (RMSEA) with 90% confidence intervals and cut-off value close to .07 (Steiger, 2007).

Following advice from Hooper, Coughlan, and Mullen (Hooper et al., 2008, Table 4), we added three indicators of goodness of fit: the Wheaton *et al.*'s relative/normed chi square with a range from 2 to 5 (Wheaton et al., 1977); the CFI with a value greater than .90; and, the non-normed fit index (NNFI) also known as the Tucker-Lewis index (TLI) with a 0.90 threshold.

If the CFA models were a poor fit to the data, items with low factor loadings (<.4) were considered for removal from the model to improve model fit. Then after removal of items with low factor loadings, modifications indices (Residual Covariance Modification Indices produced by Jamovi; >20) were explored to suggest potential changes to improve fitting. Modification indices were only considered when items belong to the same dimension. Items reallocation to other dimension where assessed with respect to the meaning. The reallocations were considered only if logic is respected.

In case of model modification, Akaike's Information Criterion (AIC) was used to compare models, rather than the likelihood ratio test because modified models are not nested. Lowest AIC value was favoured.

**For the Causal attributions scale**, as the number of present factors was unclear despite theoretical framework, an exploratory factor analysis (EFA) principal component analysis with varimax rotation was used to explore the best fit. Assumption checks were performed with Bartlett's test of sphericity which tests the hypothesis ( $H_0$ ) that the variables are unrelated therefor unsuitable for structure detection and

**Table 4.** Fit indices presentation (Hooper et al., 2008).

Indices	Presentation	Cut off	Reference
SRMR	Absolute fit indice which determine how well an <i>a priori</i> model fits the sample data. The SRMR are the square root of the difference between the residuals of the sample covariance matrix and the hypothesised covariance model.	<0.08	Hu & Bentler, 1999
RMSEA	The RMSEA tells us how well the model, with unknown but optimally chosen parameter estimates would fit the populations covariance matrix. the RMSEA favours parsimony in that it will choose the model with the lesser number of parameters.	<0.07	Steiger, 2007
relative/normed chi square	The Chi-Square value is the traditional measure for evaluating overall model fit and, assesses the magnitude of discrepancy between the sample and fitted covariances matrices. Because the Chi-Square statistic is in essence a statistical significance test it is sensitive to sample size which means that the Chi-Square statistic nearly always rejects the model when large samples are used. One example of a statistic that minimises the impact of sample size on the Model Chi-Square is Wheaton <i>et al.</i> 's relative/normed chi-square ( $\chi^2/df$ ).	[2;5]	Wheaton et al., 1977
CFI	The Comparative Fit Index is a revised form of the NFI which takes into account sample size that performs well even when sample size is small.	>0.90	Hu & Bentler, 1999
NNFI/TLI	the Non-Normed Fit Index assesses the model by comparing the $\chi^2$ value of the model to the $\chi^2$ of the null model.	>0.90	Hooper et al., 2008

Kaiser-Meyer-Olkin measure of sampling adequacy (KMO) which indicates the proportion of variance in the variables that might be caused by the underlying factors (Dziuban & Shirkey, 1974). Items loading greater than 0.4 were assigned to a specific factor. Split items between several factors were assigned to a specific factor if the square of the loading for a factor was greater than 50% that of its loading on any other factor (Wittkowski et al., 2008). When this procedure could not lead to factor assignment, the split items' contribution to the reliability of each possible subscale was examined.

**For both the Beliefs scale and the Causal attribution scale**, discriminant validity was assessed through evaluation of the intercorrelations between the factors and internal consistency was assessed using McDonald's omega, which is based on a common factor analysis model and is more accurate than the Cronbach's alpha (Béland et al., 2017).

## 2.4 Ethics statement

This study received ethical approval n°2018-A01662-53 by the French Committee for the Protection of Persons (CPP) Sud-Est II.

## 3. Results

### 3.1 Characteristics of the sample

Five hundred and seventeen participants were recruited, for a result of 406 answers (78.53%), aged 18–72 years ( $m = 39.54$ ;  $SD = 11.47$ ) with genotype SS (76.65%), SC (17.12%), or S $\beta$  (5.25%). 44.55% were born in France and 52.33% in Africa. Participants were typical of those reported in other studies presenting for SCD in a French setting (Oudin-Dogliani et al., 2019). The level of study of the participants is equivalent to that found in the general French population (INSEE, 2020): 31.23% ( $n = 158$ ) of the participants are undergraduate, 30.63% ( $n = 155$ ) have a bachelor degree and 22.33% ( $n = 113$ ) have a master degree or a PhD.

Of the 517 patients included, 514 provided information on their hydroxyurea (HU) intake. 50% ( $n = 259$ ) of patients take it, of which 94% have an SS genotype ( $n = 243$ ). Other treatments were explored, such as bleeding or blood transfusions. These data are consistent with the French prescription recommendations for HU which target more severe patients (Habibi et al., 2015).

### 3.2 The beliefs scale

The CFA assessing the original seven-factor model using all 38 items is reported as Model 1 in Table 5. Although the two-index strategy (SRMR/RMSEA) was consistent with the original factor structure, the data did not fit perfectly as indicated by the goodness-of-fit measures (CFI and TLI).

Two items, item 20 ( $\lambda = .159$ , 'My treatment will be effective in curing my sickle cell disease') and item 28 ( $\lambda = .356$ , 'I have a clear picture or understanding of my condition'), show low ( $<.40$ ) but significant loading to their respective dimensions. Item 20 was

**Table 5.** Goodness-of-fit indices for the original and revised models of the SCD-IPQ-R ( $n = 406$ ).

Cut-off	SRMR <.08	RMSEA		CFI > .90	TLI > .90	$\chi^2$ /dF			AIC Lowest
		<.07	90% CI			$\chi^2$	dF	<5	
Model 1	.077	.058	.054-.061	0.83	0.81	1516	644	2.35	42934
Model 2	.075	.058	.054-.062	0.84	0.83	1353	573	2.36	40629
Model 3	.071	.048	.044-.052	.089	.088	1100	565	1.95	40392
Model 4	.058	.042	.038-.047	.092	.091	973.5	562	1.73	40272

purported to measure Cure and item 28, Coherence. All other items loading were significant and sufficient, ranging from .40 to .847.

For a modified model (Model 2), we dropped the above-mentioned items with poor loadings and conducted CFA on the remaining 36 items. Goodness-of-fit indices suggested an improvement of the model as compared to the initial model, especially given that SRMR/RMSEA were still in line with cut-off criteria. However, CFI and TLI were still not in line within level of acceptable fit. Eight modification indices ( $MI > 20$ ) suggested that relevant modifications could be made in the model.

These modifications were carried out in a third model (Model 3) and implemented one by one. Error covariances were added between:

- Items 21 ('The negative effects of my sickle cell disease can be prevented (avoided) by my treatment') and 22 ('My treatment can control my sickle cell disease') ( $MI = 53.92$ ) belonging to Treatment control/Cure, items 15 ('Nothing I do will affect my sickle cell disease') and 17 ('My actions will have no effect on the outcome of my sickle cell disease') ( $MI = 36.97$ ) belonging to Personal control, items 34 ('When I think about my sickle cell disease I get upset') and 38 ('My sickle cell disease makes me feel afraid') ( $MI = 28.88$ ) belonging to Emotional representation.
- Items 37 ('Having sickle cell disease makes me feel anxious') and 38 ('My sickle cell disease makes me feel afraid') ( $MI = 34.14$ ) belonging to Emotional representation.
- Item 6 ('My sickle cell disease is a serious condition') and 7 ('My sickle cell disease have major consequences on my life') ( $MI = 29.79$ ) belonging to Consequences.
- Items 14 ('The course of my sickle cell disease depends on me') and 16 ('I have the power to influence my sickle cell disease') ( $MI = 25.16$ ) belonging to Personal Control.
- Items 1 ('My sickle cell disease will last a short time') and 4 ('This sickle cell disease will pass quickly') ( $MI = 24.56$ ) belonging to Chronology.
- And items 23 ('There is nothing which can help my condition') and 24 ('The symptoms of my sickle cell disease are puzzling to me') ( $MI = 21.51$ ) belonging to Coherence.

The re-specified model resulted in a considerably better fit: SRMR/RMSEA were still under their specific cut-off, and CFI or TLI were very close to .90.

Items reallocations to other dimensions were performed (Model 4) and implemented one by one:

- Item 24 ('The symptoms of my sickle cell disease are puzzling to me') ( $MI = 71.39$ ) is associated with Emotional Representation.

- Item 18 ('My sickle cell disease will improve in time') (MI = 24.51) is associated with Treatment control/Cure.
- Item 36 ('My sickle cell disease does not worry me') (MI=) is associated with Consequences.

This final model verifies the necessary goodness-of-fit meaning that the final model fit the data well.

Internal consistencies of the seven dimensions of the modified 36-item Beliefs scale (items 20 and 28 dropped) is good (Emotional representation, six items,  $\omega=.87$ ; Timeline-acute-chronic, six items,  $\omega=.83$ ; Consequence, six items,  $\omega=.80$ ; Coherence, four items,  $\omega=.76$ ; Timeline-cyclical, four items,  $\omega=.73$ ) through marginally questionable (Personal control, six items,  $\omega=.67$  and Cure, four items,  $\omega=.61$ ).

Correlation between the seven-core latent factors shows weak intercorrelations except for the correlation between Consequence and Emotional representation ( $r = .58$ ;  $p < .001$ ) suggesting that these two dimensions are moderately correlated.

### 3.3 The causal attribution scale

The thirty-two causal items are suitable for EFA (Bartlett's Test of Sphericity  $\chi^2(496) = 3383$ ;  $p < .001$  and KMO = .821). The EFA results (based on parallel analysis, with a maximum likelihood extraction completed by a varimax rotation) supported a 6-factor structure with only 19 items reaching a loading higher than 0.40. The overall variance was low at 38.09%, but fitting measures suggested a somewhat acceptable fit for an EFA (RMSEA = .055 [.050; .062]; TLI = .082;  $\chi^2/Df = 2.03$ ).

The first two factors share half of their items, suggesting that they refer to a single dimension, the perceived risk factors (43, 44, 49, 55, 61, 62, 67, 68, and 69). The third factor comprises two items (48 and 54) dealing with religion. The fourth factor is composed of two items (65 and 66) associated with alcohol and smoking, suggesting healthy behaviours. The fifth factor groups together four items of which, three are positively correlated (47, 51, 57) referring to heredity, and one negatively correlated (50) referring to microbes or virus. This fifth factor suggests a good understanding of what causes sickle cell disease. Finally, the sixth factor referred to Africa with two items (40 and 46).

Internal consistency of the final five factors is correct (Factor 1: Risk factors, nine items,  $\omega=.859$ ; Factor 2: Religion, two items,  $\omega=.817$ ; Factor 3: Healthy way of life, two items,  $\omega=.865$ ) through marginally questionable (Factor 4: Heredity, four items,  $\omega=.634$  and Factor 5: Africa, two items,  $\omega=.542$ ).

Intercorrelation matrix show no significant correlations suggesting that these dimensions do not refer to the same causal attribution.

## 4. Discussion

Sickle cell disease, a genetic condition, is characterised by severe pain and has a high impact on the quality of life and the psychological well-being of patients. Numerous studies have shown that these psychological factors influence the health status of patients. In other chronic illnesses, patients' self-perception of their illness has been shown to influence their health outcome. The work presented here aimed at examining the

hypothesised factor structure of the IPQ-R among a population of adults with sickle cell disease. Our findings suggest that although the hypothesised factor structure provides a good fit to the data, this fit can be improved by making three adjustments to item allocation:

- Item 24 appears to be more related to the Emotional representation dimension than the Coherence factor. Item 24 assesses the perception of being puzzled by the symptoms of sickle-cell disease. The other items of the Coherence dimension refer to cognitive mechanisms like ‘understanding’, ‘making sense’, or ‘having a clear picture’ and do not refer to perplexity. Perhaps the reference of ‘puzzled’ made by item 24 provokes feelings about the symptoms manifestation rather than cognitions.
- Item 18 seems more related to the Treatment control factor than the Timeline-acute/chronic factor. Item 18 assesses the conviction that sickle cell disease will ‘improve’. The other items on the Timeline-acute/chronic factor refer to the duration of the disease and not to the improvement of the condition. Reference to improvement could elicit cognition about the possible controllability of the disease. Dempster and McCorry (2012) hypothesised that item 18 is better associated with Treatment control, due to its placement in the questionnaire before the other Treatment control factor items. Other psychometric validations found the same issue with item 18, suggesting that it should either be placed with the other Timeline-acute/chronic items or completely removed (Chen et al., 2008; Giannousi et al., 2010).
- Finally, item 36 gives the impression of being more related to the Consequence factor than the Emotional representation factor. Item 36 assesses the belief that sickle cell disease ‘does not worry’ the respondent. The other item of the Emotional representation factor refers to specific feelings or psychopathology like depression (item 33), distress (item 34), anger (item 35), anxiety (item 37), or fear (item 38). The factor summarising emotional representation is composed of items referring to emotions, and does not refer to cognitive processes, such as concerns. The emotions or feelings refer to the present state of the patient while concerns are an anticipation of the future. We could hypothesise that apprehension of sickle cell disease is linked to the consequences the condition has on daily life.

Secondly, eight pairs of items share a significant proportion of their variance that is not explained by their respective factor.

- In the Treatment control dimension, items 21 and 22 refer to the beliefs that ‘the negative effects of sickle cell disease can be prevented by treatments’ and that ‘treatment can control sickle cell disease’, respectively. In the Cure dimension, since item 20 has been removed due to low factor loading, the two remaining items (19 and 23) do not refer to treatment, but to beliefs that ‘little can be done to improve my sickle cell disease’ or that ‘there is nothing which can help my condition’. The items in the Treatment control factor tend to divide into, on the one hand, a set of items dealing with the control of the disease by the treatment with items 20, 21 and 22, and on the other hand, a set of items referring to helplessness with items 19 and 23 to which item 18 could be added. Splitting the Treatment control dimension into two dimensions, Treatment control and Sens of helplessness, should be considered.

- Considering the Personal control factor, items 14 and 16, and items 15 and 17 share an important amount of their variance. Items 14 and 16 refer to the positive beliefs that ‘the course of sickle cell disease depends on’ the respondents and that they have ‘the power to influence’ their condition. On the contrary, items 15 and 17 relate to negative cognitions, including ‘nothing could affect the disease’ or ‘actions will have no effect on the outcome of the disease’. These two pairs denote positive and negative aspect of personal control. However, as items 15 and 17 have reverse scoring, the balance between positive beliefs and negative cognition is cancelled.
- For the Emotional representation dimension, two pairs of items seem to share some variance, items 34 – 38 and 37 – 38. These items make reference to distress, anxiety and fear, which appears to be a different facet of the anxiety spectrum (Clark & Watson, 2006; McTeague & Lang, 2012).
- In the Consequence dimension, items 6 and 7 make reference to the personal negative consequences while the other items refer to how others feel or are impacted by the disease. The items show a tendency to divide into personal consequences and consequences for others.
- Finally, in the Timeline-acute/chronic dimension, items 1 and 4 refer to the belief that sickle cell disease ‘will last a short time’ and ‘will pass quickly’, respectively. The remaining items refer to the belief that the condition ‘is likely to be permanent rather than temporary’, ‘will last for a long time’ and will be present ‘for the rest of my life’. Therefore, the items show a tendency to divide into acute and chronic timelines. This division indicates that, to some extent, people with sickle cell disease hold on to the belief that their disease is both permanent and temporary: it has a genetic origin, but it is also temporal, as there may be a confusion between painful episodes and the whole disease itself (Oudin-Doglioni et al., 2019). This specific perception of temporality in SCD and its variations may be influenced by other covariates such as healthcare utilisation or age.

Consequence and Emotional representation show mild correlation. This correlation has been found in most studies examining the psychometric proprieties of the IPQ-R and will not be further discussed.

Compared to the nine other studies that performed the same work as us, the comparison is complicated since the adaptation of the IPQ-R is dependent on the population in which it was completed. In order to achieve sufficient goodness-of-fit indices, modifications made by other authors were based on their data. The question therefore arises of the comparability of the results obtained with the IPQ-R between different populations of patients. What appears certain is that the underlying structure of the IPQ-R is found in all the studies, in particular the seven dimensions of the Beliefs scale, before any modifications to improve the fit to the data. Therefore, the raw results of the IPQ-R can be compared, since they refer to the same dimensions. However, the operations of item deletions or reallocation of items in other dimensions modify the structure of the dimensions, which prevents a comparison between the different populations if the adaptation does not contain the same structural modifications. Thus, before making any comparisons between different patient populations, we must ensure that the structure of the dimensions of the IPQ-R are comparable, i.e. either the use of raw data or that the IPQ-R undergoes the same modifications (deletion and/or reallocation).

The Leventhal's model assumes that health behaviour can be explained and predicted from patients' representations of their disease. In this sense, the adaptation of IPQ-R to adults with sickle cell disease provides us with a theoretical framework for understanding the adoption of health behaviour. In particular, the issue of treatment adherence in this population is particularly important. Several studies have already shown that adherence is very low in adults with SCD (Badawy et al., 2017; Oudin-Doglionni et al., [Under editorial process](#)). Applying the Leventhal's model to this specific question would make it possible to highlight the particular representations of adults with SCD and then, in a second step, to propose specific interventions to improve their adherence.

Of the 32 items included in the causal attribution scale, 19 reach sufficient factor loading, which is divided into a five-factor model. A first factor referring to risk factors such as psychological risk or risk link to the way of life. The second factor refers to magico-religious factor. The third deals with healthy life without alcohol or tobacco, the fourth with heredity, and the fifth links sickle cell disease to Africa. All these factor have a relation with the anthropological and ethnographical studies (Gernet et al., 2011; Lainé, 2004). All the items included in the original version were not found in the final Causal attribution scale, confirming that perceived cause of a disease is dependent on the illness. This scale is very specific to our population. According to our findings, researchers using IPQ-R for other diseases, especially those for which it is not yet validated, should pay special attention to this scale.

The results were obtained on a sample of just over four hundred adults, which is less than the average inclusion of other studies examining the psychometric properties of the IPQ-R, close to six hundred inclusions. A reproduction of this study on a larger sample could confirm these initial results. Moreover, this study was carried out in the National Reference Centre for Sickle Cell Disease, which could possibly represent a selection bias. The excellent care in the centre can influence the perception that patients have of sickle cell disease. Opening this questionnaire to other hospitals and SCD care centres could alleviate those doubts. However, this exploration of the psychometric properties of IPQ-R is the first to have been carried out on a population suffering from a genetic disease. It highlighted several problems concerning the allocation of certain items and the presence of particularly strong interitem correlations. In this sense, more general work on the structure of the dimensions should be considered since several of the studies that have explored the psychometric properties of the IPQ-R have reported the same concerns. Finally, none of the studies that have explored the psychometric properties of the IPQ-R has been able to ignore structural modifications to obtain a good match with the fit indices.

This recurrence confirms the need for new work on the IPQ-R, like that carried out by Moss-Morris (Moss-Morris et al., 2002), which would allow an ambitious restructuring of all dimensions. Especially since a meta-analysis of 23 datasets from 30 studies using IPQ-R in chronically ill patients (Brandes & Mullan, 2014) indicates moderate to high heterogeneity for all dimensions apart from timeline, coherence and emotional representations, with funnel plots indicative of bias. In a context of reproducibility issue (Wingen et al., 2019), the robustness of the CSM to meta-analytic investigations should be acquired (Doyle & Mullan, 2017).

This work on the psychometric properties of IPQ-R in an adult population suffering from sickle cell disease has made it possible to adapt its use to this population after the

adoption of data-driven modifications. The data are specific to the population in which they were obtained and cannot lead to a generalisation in other chronic pathologies even of genetic origin. This research, like other studies that have explored the psychometric properties of the IPQ-R, is an invitation to work on a larger scale on the structure of the dimensions of this tool. Thanks to the validation made, the study of the representations of adults with sickle cell disease in France was able to be carried out and it will be able to guide the implementation of specific interventions aimed at improving their quality of life and their mental well-being.

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### Data availability statement

The data that support the findings of this study are openly available in Open Science Framework (OSF): <https://doi.org/10.17605/OSF.IO/YABFV>

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