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The PrU: development and validation of a measure to assess personal utility of genomic results

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The PrU: development and validation of a measure to assess personal utility of genomic results

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Abstract

Purpose: People report experiencing value from learning genomic results even in the absence of clinically actionable information. Such personal utility has emerged as a key concept in genomic medicine. The lack of a validated patient-reported outcome measure of personal utility has impeded the ability to assess this concept among those receiving genomic results and evaluate the patient-perceived value of genomics. We aimed to construct and psychometrically evaluate a scale to measure personal utility of genomic results – the PrU.

Methods: We used an evidence-based, operational definition of personal utility, with data from a systematic literature review and Delphi survey to build a novel scale. Following piloting with 24 adults, the PrU was administered to healthy adults in a Clinical Sequencing Evidence-Generating Research Consortium (CSER) study after receiving results. We investigated responses using exploratory factor analysis.

Results: The exploratory factor analysis (*n*=841 participants) resulted in a three-factor solution, accounting for 74% of the variance in items: 1) self-knowledge (α=.92), 2) reproductive planning (α=.89), and 3) practical benefits (α=.91).

Conclusions: Our findings support the use of the three-factor PrU to assess personal utility of genomic results. Validation of the PrU in other samples will be important for more wide-spread application.

Key words: Patient Reported Outcome Measure; Psychometrics; Health Services Evaluation; genetic counseling; bioethics; ELSI; Perceived Value

Introduction

Utility in medicine is a measure of the level of benefit resulting from an intervention or procedure.[1] Such utility is commonly conceptualized in terms of clinical benefit – clinical utility – the likelihood of an improved health outcome based on evidence-informed recommendations for follow-up.[2] Clinical utility in the most narrow sense is an objective measure of health status.[3]

Genomic tests do not in themselves have clinical utility; rather, these tests provide information, which may in turn be used to indirectly improve health.[4] The ability of genomic information to lead to improved health outcomes may be limited for three primary reasons.[5] First, there may be inadequate data to determine the pathogenicity of a variant and therefore the effect of the genomic information on disease risk is uncertain.[6] Second, there may not be a proven medical intervention to mitigate disease.[7] Third, genomic information may not be communicated clearly, in a patient-centered way.[8] Yet, people receiving genomic information report benefits even in the absence of clinical utility.[9, 10]

Personal utility is a patient-centered construct based on evidence of non-clinical benefits in genomic medicine. The construct includes thoughts, feelings and behaviors related to undergoing genome sequencing and genetic testing.[9, 11] Prior empirical work has resulted in a definition of personal utility as non-medical perceived benefits, such as increased self-knowledge, knowledge about the condition, altruism and coping.[9, 11]

The clinical and personal utility of a diagnostic test is of central importance to making medical decisions about whether to pursue testing.[4] The application of genomic medicine has been variable, with inconsistencies in insurance coverage and disparities in access, particularly among historically underrepresented groups.[12] Addressing such inconsistencies relies on the ability to accurately measure and assess genomic medicine outcomes, including both clinical and personal utility. Efforts have led to the development of a clinician-reported measure of clinical utility.[13] While there is increasing recognition of the importance of assessing non-clinical

benefits,[4, 5, 10] there is no validated patient-reported scale to measure personal utility in genomic medicine.

The ability to assess the role of specific elements of personal utility will enhance our understanding of the outcomes experienced by participants and patients who undergo genomic testing and guide optimal translation in clinical care, including assessment of the overall value of genomic medicine. We aimed to develop and validate a novel scale to measure the personal utility of genomic results (the PrU).

Materials and Methods

Initial Scale Development

We (Kohler, Turbitt and Biesecker) previously identified elements of personal utility in genomic medicine through a systematic literature review and used these elements to assemble a scale including 35 items representing 15 elements of personal utility.[9] We refined these items through a modified Delphi assessment involving adult participants of a genomic sequencing study.[11] For the Delphi, participants were asked to rate each of the 35 items of personal utility as an outcome from learning sequencing results, first for themselves and next for others. Ratings were indicated on a Likert-type scale assessing how plausible each item was perceived to be. Two rounds of surveys were administered to the same participants. After each round items lacking endorsement were removed. After both rounds, 24 items representing personal utility remained.[11]

To assemble the PrU we phrased the 24 items as statements with the stem *"Please indicate how useful you find the following outcomes of your test result"* and each statement could be rated on a 7-point, Likert-type scale ranging 1=Not at all Useful to 7=Extremely Useful according to how useful the item was perceived to be as an outcome of genomic testing. For example, an end-user could rank the item "help me or my family mentally prepare for the future". The wording of the statements was modified slightly to create two versions of the PrU; a parent

version for adults receiving results from their child's genomic test and adults receiving results about themselves. The adult version of the PrU is the focus of the analysis reported here.

Piloting and scale refinement

We piloted the scale with 24 healthy adults who had undergone genome sequencing (19 over-the-phone, 5 online). Those who participated in online piloting (using the platform SurveyMonkey) were recruited from the Medseq study at Brigham and Women's Hospital and Harvard Medical School, Boston.[14] Over-the-phone piloting was carried out with ClinSeq® participants.[15] The online piloting process aligned with the format for which the scale was intended to be used, while over-the-phone piloting enabled collection of more comprehensive verbal feedback.

We presented participants with the scale items for piloting, as well as brief introductory instructions asking people what was important to them about receiving results from genomic testing (see Box S1 in supplemental information for details). We asked participants to rate each item using the 7-point Likert-type scale described above, describe what the item was referring to in their own words (open text), and whether they thought the item should be included in a study exploring personal utility (yes, no, maybe). For piloting over-the-phone, participants were also asked whether they would "ask the question differently" (open response). Open text data from the pilot were analyzed using content analysis and indicated changes were discussed with the research team.

We developed an interim scale after piloting where we reduced the number of items from 24 to 19 as unclear and redundant items were identified by participants and agreed upon by the research team. For example, the item "help me live more fully" was dropped as pilot participants had difficulty interpreting the meaning and the research team determined that the item overlapped with many of the other scale items.

The Clinical Sequencing Evidence-Generating Research (CSER) consortium is a collection of seven US federally funded programs of research that seek to provide evidence about the effectiveness of implementing genome sequencing into clinical care. CSER research projects used survey measures harmonized across the research programs.[16] No validated scale to measure personal utility in genomic medicine was available when CSER projects commenced, and thus this presented an opportunity for gathering validation evidence for the novel scale. The scale was shared with CSER investigators for inclusion in project surveys. CSER investigators suggested the removal of two further items (from 19 to 17 items) that represented overlapping concepts. The adult version with a total of 17 items administered for this study is in Table 1.

Sample and data collection for exploratory factor analysis

The 17-item scale was administered to CSER participants at follow-up, 0–4 weeks after disclosure of genomic results. Two sites (Clinseq® and CHARM) administered the adult version of the scale. Eligibility criteria for the Clinseq® study included self-identifying as African-descended, 45-65 years old, not having smoked in the past year, living in the Washington, DC area, and not enrolled in another sequencing study returning individual results. Eligibility for the CHARM study included being a Kaiser Permanente Northwest or Denver Health patient, screening high risk for a hereditary cancer syndrome (or an unknown family history), no prior testing for cancer predisposition variants associated with Lynch syndrome or hereditary breast and ovarian cancer, and English or Spanish speaker.

Potential clinical benefits were discussed with participants. For example, during the informed consent process, individuals were told that their genomic results could change the medical care their doctor recommends. Concepts measured in the PrU scale were not routinely discussed with participants. Most participants from both sites received negative reports. In the CHARM study, 5% had a pathogenic or likely pathogenic finding and 9% had a variant of

uncertain significance in a cancer risk gene. A further 1% had a secondary finding.[17] Among Clinseq® participants, 2.6% received a secondary finding.[18]

Descriptive variables included demographic characteristics (age of participant, sex, education, and race), self-report general health (excellent, very good, good, fair, or poor), and weeks post return of results the scale was completed. We assessed convergent validity using scores from the positive feelings subscale of the FACToR (Feelings About genomiC Testing Results) scale.[19] FACToR is a 12-item validated scale that measures the psychological impact of genomic results. The positive feelings subscale consists of the following 4 items: How happy did you feel about your genetic test result?; How relieved did you feel about your genetic test result?; How much did you feel that you understood clearly your choices for disease prevention or early detection?; How helpful was the information you received from your genetic test result in planning for the future?. Response options range from 4=not at all, to 0=a great deal (note that FACToR measures negative psychological impact, we reversed the sign to facilitate interpretation of the reverse scoring). The FACToR scale was only administered by one of the two CSER sites used in this analysis. We expected that participants who reported high personal utility scores would also report higher positive feelings.

Statistical analysis

We began by examining the descriptive statistics for the items to determine whether there were any strong floor or ceiling effects or differential non-response. We used exploratory factor analysis to examine the structural validity of the 17 items that are purported to measure personal utility. We evaluated the factorability of the items using the Kaiser–Meyer–Olkin (KMO) test, requiring a value of at least .70 to be considered adequate.[20] The KMO Measure of Sampling Adequacy is a statistic that indicates the proportion of variance in items that might be caused by underlying factors, i.e., the proportion of variance across all of the items that is attributed to common variance. This, along with the Bartlett Test of Sphericity, are the most

common indices used to determine whether a set of items or variables is appropriate for factor analysis.

We extracted the factors using principal-axis factoring, which focuses on the common variance among the items and used direct oblimin rotation to facilitate interpretation of the extracted factors while allowing for a correlation among the factors using the pattern matrix. We determined the number of factors to extract based on eigenvalues, scree plot, and theoretical sensibility of candidate factor solutions. That is, a solution that explains a substantial amount of variance that also produces a logical pattern with simple structure.[21]

We examined the internal consistency using Cronbach's alpha for each factor. We took the mean of all items to create an overall PrU score and items in each factor to create subscale scores. We evaluated convergent validity by examining the association between the positive feelings subscale of the FACToR and PrU overall and subscale scores using Pearson's correlation.

Results

Respondent characteristics

In total, 900 participants across two CSER sites provided responses to the adult version of the PrU. There were 59 participants who did not provide responses to all PrU items and were removed from the analysis, leaving 841 valid responses (Table 2). The mean number of missing responses per item was 7/900 (0.8%) and ranged from 4 to 19 (item 4: Use for testing a future pregnancy, if appropriate). Because 841/900 (93.4%) had complete data on all of the items and there was no apparent pattern that we could discern, we used listwise deletion/complete case analysis. This approach was appropriate given that the required analytical effort to perform an appropriate multiple imputation would likely not produce substantively different results from a listwise approach.[22]

Responders were mostly female (*n*=599, 77.2%), educated to some post-high school or beyond (*n*=571, 73.6%) and identified as White (*n*=332, 37.7%), Black (*n*=251, 28.5%) and/or Hispanic/Latino (*n*=188, 21.3%). The average age of responders was 41.7 years (*SD*=11.9). Most were in good, very good, or excellent health (*n*=636, 80%). Responses were provided on average 2.1 weeks after receiving genomic results (*SD*=2.0), ranging from 0 weeks to 27 weeks.

Exploratory factor analysis results

An examination of the items revealed no evidence of strong ceiling or floor effects, nor did any of the items appear to be more likely to be missing. Table S1 of supplemental information provides the mean, median, standard deviation, skewness and kurtosis of each item. The KMO test across the items was .94, indicating that the items share a great deal of common variance and would be appropriate for factor analysis. Bartlett's test of sphericity was significant ($\chi 2$ (136) = 11224, p<.001).

Based on the eigenvalues and the scree plot (Figure S1 of supplemental information), we further investigated the feasibility of a two and three factor solution by examining the loadings from the pattern matrix. After consideration, we settled on a three-factor solution that accounted for 66% of the variance in the items.

We assessed items that cross-loaded as candidates for removal in an iterative process (i.e., removing a candidate item then rerunning the factor analysis; Table S2). We decided that PrU2 "inform my plans for school or career" was the top candidate for removal due to low loading. Further, the item was originally adapted from evidence among parents and is less relevant to an adult cohort.

Next, the cross-loading of PrU9 "help me feel more in control of my health" may be the result of this item sharing much of the same stem with PrU10 "help me feel more in control of my life". We determined it was more important to retain PrU10 as it aligns with our definition of

personal utility as non-health outcomes more closely than PrU9. PrU10 no longer cross-loaded once PrU9 was removed.

Lastly, we decided to remove PrU17 “feel good about taking responsibility for my health”, due to low loading. This item was adapted from literature about parents’ feelings of responsibility for passing on genetic risks to their children. We suspected this item did not load well because it resonated more with the parent sample and does not appear to substantively add to any of the factors compared to the remaining items.

The new three-factor solution with 14 items accounted for 74% of the variance in the items. The first factor accounted for 55% of the variance and items that clearly loaded on this factor were centered around self-knowledge. The second factor accounted for an additional 12% of the variance and consisted of two items that were concerned with reproductive planning. The third factor accounted for an additional 7% of the variance and consisted of items that dealt with practical benefits (Table 3).

Overall score, internal and external consistency

The Cronbach’s *α* for all 14 items was .93. We used the mean of the 14 items to generate a total score, and for this sample the mean was 4.8 (*SD*=1.2; response range was from 1 to 7), representing a slightly higher than neutral response. The Cronbach’s *α* and descriptive statistics of the PrU subscales (computed by taking the mean of the items that comprise each assigned factor) were: self-knowledge, *α*=.92, *mean*=5.5, *SD*=1.2; reproductive planning, *α*=.89, *mean*=3.4, *SD*=2.0; practical benefits, *α*=.91, *mean*=4.6, *SD*=1.5. See Figure S2A-D of supplemental information for histograms of overall score and subscale response frequencies.

There was a positive association between the FACToR positive feelings subscale and PrU overall score, which were correlated (*r*= .61, *p*<.001, *N*=594). As expected, those with higher PrU scores also had higher positive feelings about their genetic test results. Each of the

subscales were positively correlated with the FACToR positive feeling subscale: self-knowledge (r= .58, p<.001), reproductive planning (r= .34, p<.001), and practical benefits (r= .57, p<.001).

Discussion

Our report of the PrU is the first attempt to develop and validate an evidence-based scale to measure personal utility concepts that are most relevant to end users of genomic testing. Our findings offer a useful starting point for future efforts to refine and validate the scale. Our reliability and validity data provide proof-of-concept that it is possible to measure personal utility in genomic medicine. The three factors that emerged from the current study are centered on: self-knowledge, reproductive planning, and practical benefits.

'Self-knowledge' represent the benefits of learning more about oneself from undergoing genetic testing. These include cognitive aspects, such as increased understanding of one's genome, as well as affective aspects such as positive feelings related to the knowledge gained helping others. By contrast, the other two factors reflect pragmatic applications of genetic testing information. In 'practical benefits', outcomes relate to future planning, access to programs, and communication with family members. In 'reproductive planning', these are specific to family planning. We consider the reproductive planning items a component of personal utility as use of test results in this way does not lead directly to improved health outcomes for the individual.

The subscales were positively correlated with the FACToR positive feelings subscale whereby those reporting higher personal utility also felt more positive about their results. The association was weaker between FACToR positive feelings and the PrU reproductive planning subscale compared to the other two PrU subscales. This weaker association is likely due to the influence of life stage on the PrU reproductive planning subscale. Many respondents were post-reproductive age.

There were differences between the three-factor structure reported here and our previously defined domains of personal utility resulting from a systematic literature review.[9] Our

review generated four domains of personal utility: affective, cognitive, behavioral and social outcomes. There is overlap between the literature review domains and factors described here; for example, 'practical benefits' aligns with the behavioral domain, as many of the items in the practical benefits factor involve expected behavioral outcomes such as use of social programs and life planning. However, the factor analysis results from our current study suggest that the pattern of responses from users are driven by more specific and descriptive concepts of personal utility.

The value of genomic medicine has been discussed broadly in the literature, however, there remains no widely accepted approach to assess its utility. Diverse stakeholder groups and domain experts each conceptualize utility through a specific lens.[10] From the medical perspective, value is traditionally assessed as clinical utility, or how likely an intervention will have clinical benefit or inform management; from the payor perspective, focus is placed on cost-effectiveness often measured through cost-utility analyses; from the patient perspective, utility of genomic medicine spans clinical outcomes as well as emotional and informational benefits.[10, 23, 24] Recognition of personal utility as a key outcome of genomics enables further investigation into how this concept is associated with health outcomes, which may lead to more holistic frameworks to assess a person's health.

Our efforts toward measuring and assessing patient-perceived personal utility in genomic medicine align with a wider transformation in medicine to meaningfully engage patients in determining outcomes most important to them.[25] While further validation of the PrU is necessary, we predict a range of potential applications. Use of the PrU can enable clinicians or researchers to anticipate what benefits their patients may experience from genomic testing and inform discussions around test decision making. Researchers assessing the value of genomic medicine in a specific context, or the effectiveness of novel interventions and protocols can use the PrU as an outcome measure. Such evidence may contribute to health technology assessments of genomic medicine and related products.[26]

Strengths and limitations

A key strength of the PrU is the rigorous, evidence-based methodology used in its development. Further, the identified constructs produced by the factor analysis make sense in context of empirical and theoretical evidence. Our sample was ethnically/racially diverse.

There are limitations to our study that should be noted. Our sample is more highly educated compared to the general population. This may limit generalizability of the scale for use in studies with individuals with lower literacy and future work should test and validate the scale among a population that includes such individuals. For example, further work could be carried out to adapt the scale to an easy-read format to be administered to individuals with intellectual disability undergoing genome sequencing.[27] The scale was developed specifically for use in the CSER consortium studies, with some pragmatic decisions made in developing the scale for use in this context including the removal of items to reduce participant burden. Further testing of the scale in other contexts and time-points is important to determine that the full spectrum of personal utility is captured in the scale and to determine changes in personal utility over time.

We were unable to link participant responses to the PrU with the variant type returned. However, the majority of participants received negative findings and the small proportion of those who received a pathogenic finding are unlikely to impact our validation results. Further work to validate the PrU among individuals receiving result types other than negative is required.

Finally, while we intended to measure experienced utility, the 4-week timeframe for participants to provide responses may mean that not all aspects of utility were experienced. In this case, participants may have provided responses regarding expected or future utility. This may be an important consideration for use of the scale to measure experienced utility. We do not expect validation analysis to be affected.

Conclusions

We provide initial psychometric properties of the novel PrU; a patient-reported outcome measure of personal utility in genomic medicine. Our results suggest strong evidence for three subscales of personal utility valued by adults that converge on the broader concept of personal utility. It is crucial that efforts to measure personal utility continue as it is a highly variable and individualized concept. The ability to measure the dimensions of utility in genomic medicine contributes to understanding patient experiences and guiding the implementation of genomics in clinical care.

Data availability: Data are available on request.

Author contributions: Conceptualization: E.T., J.N.K., F.A., B.B.B., M.C.L.; Data curation: F.A., M.C.L.; Formal analysis: M.C.L.; Investigation: E.T., J.N.K., F.A., I.M.M., K.L.L., K.A.B.G, B.S.W.; Resources: K.A.B.G., B.B.B.; Software: M.C.L.; Visualization: E.T., M.C.L: Writing-original draft: E.T., J.N.K.; Writing-review & editing: E.T., K.N.K., F.A., I.M.M., K.L.L., K.A.B.G., B.S.W., B.B.B., M.C.L.

Ethics declaration: The study protocols to collect data analyzed in this report were approved by the National Human Genome Research Institute Institutional Review Board and Kaiser Permanente Northwest Institutional Review Board. Informed consent was obtained from all participants.

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Table 1. Final version of the Personal Utility (PrU) scale

PERSONAL UTILITY SCALE (PrU) - Adult

Set survey to randomize items to avoid order effects

Please indicate how useful you find the following outcomes of your test result:

	Not at all useful	A little useful	Somewhat useful	Neutral	Useful	Very useful	Extremely useful
Help with life planning	1	2	3	4	5	6	7
Inform my plans for school or career	1	2	3	4	5	6	7
Inform my decisions about having children	1	2	3	4	5	6	7
Use for testing a future pregnancy, if appropriate	1	2	3	4	5	6	7
Help me or my family mentally prepare for the future	1	2	3	4	5	6	7
Help to better understand my health	1	2	3	4	5	6	7

Contribute to my self-knowledge	1	2	3	4	5	6	7
Help me cope with my health risks	1	2	3	4	5	6	7
Help me feel more in control of my health	1	2	3	4	5	6	7
Help me feel more in control of my life	1	2	3	4	5	6	7
Simply to provide information	1	2	3	4	5	6	7
Satisfy my curiosity	1	2	3	4	5	6	7
Help me to use social programs, like resources and services	1	2	3	4	5	6	7
Improve communication with my family members	1	2	3	4	5	6	7
Feel good about helping the medical community	1	2	3	4	5	6	7

Feel good about having information for my family members	1	2	3	4	5	6	7
Feel good about taking responsibility for my children's health	1	2	3	4	5	6	7

Table 2. PrU adult descriptive statistics *N*=841

Variable	Frequency	Proportion
Site		
CHARM (Kaiser Foundation Research Institute)	594	70.6%
Clinseq® (National Human Genome Research Institute)	247	29.4%
Age: *Mean*=41.7, *SD*=11.9, *min*=18, *max*=69		
Sex		
Female	599	77.2%
Male	168	21.6%
Transgender	4	0.5%
Does not identify as F, M, or T	5	0.6%
Education		
Bachelor's degree (for example: BA, AB, BS)	215	27.7%
Graduate or professional degree (for example: MA, MBA, JD, MD, PhD)	198	25.5%

	Some post-high school training (college or occupational, technical, or vocational training), no degree or certificate	159	20.5%
	Associate (2-year) college degree, or completed occupational, technical, or vocational program and received degree or certificate	79	10.2%
	High school graduate (diploma or GED equivalent	61	7.9%
	Less than high school (less than 9th grade)	45	5.8%
	Some high school (9th to 12th grade), no diploma	20	2.6%
Race			
	White or European American	332	37.7%
	Black or African American	251	28.5%
	Hispanic/Latino(a)	188	21.3%
	Asian	40	4.5%
	American Indian, Native American, Alaska Native	39	4.4%
	Native Hawaiian/Pacific Islander	11	1.2%

Unknown/none of these fully describe me	11	1.2%
Middle Eastern of North African/Mediterranean	5	0.6%
Prefer not to answer	4	0.5%

Weeks post return of results scale completed: *Mean*=2.1, *SD*=2.0, *min*=0, *max*=27

General Health

Excellent	56	7.0%
Very good	269	33.8%
Good	311	39.1%
Fair	144	18.2%
Poor	15	1.9%

Some variables do not sum to 841 due to missing data

Participants could select more than one Race category

Table 3. Final factor structure and factor loadings

	Factors		
PrU items	**1**	**2**	**3**
PRU11 Simply to provide information	.933		
PRU7 Contribute to my self-knowledge	.827		
PRU12 Satisfy my curiosity	.827		
PRU6 Help to better understand my health	.694		
PRU15 Feel good about helping the medical community	.594		
PRU16 Feel good about having information for family members	.571		-.326
PRU3 Inform my decisions about having children		.913	
PRU4 Use for testing a future pregnancy, if appropriate		.882	
PRU14 Improve communication with my family members			-.855
PRU13 Help me to use social programs, like resources and services			-.748
PRU1 Help with life planning			-.592
PRU10 Help me feel more in control of my life			-.568

PRU8 Help me cope with my health risks	.333	-.558
PRU5 Help me or our family mentally prepare for the future		-.548

The PrU: development and validation of a measure to assess personal utility of genomic results

Supplemental information

Box S1. Introductory instructions for scale pilot participants

> We are asking people about what is important to them about receiving results from sequencing [...] We will use the information to improve the items we are asking about to make sure they are well worded and convey what we hope [...] I am asking you to go through reasons why some people are interested in learning their genetic information. We know that people are often interested in learning medical information from genetic test results, such as learning about health risks. But we also have learned that people have other non- medical reasons for being interested in learning their genetic information.
>
> We have developed a way to survey people about non-medical reasons they may have for wanting to learn their genetic information. But we have not used these survey items yet because we need help in making sure they mean what we intend.

Figure S1. Scree plot for exploratory factor analysis indicating the 3 factor structure of the PrU

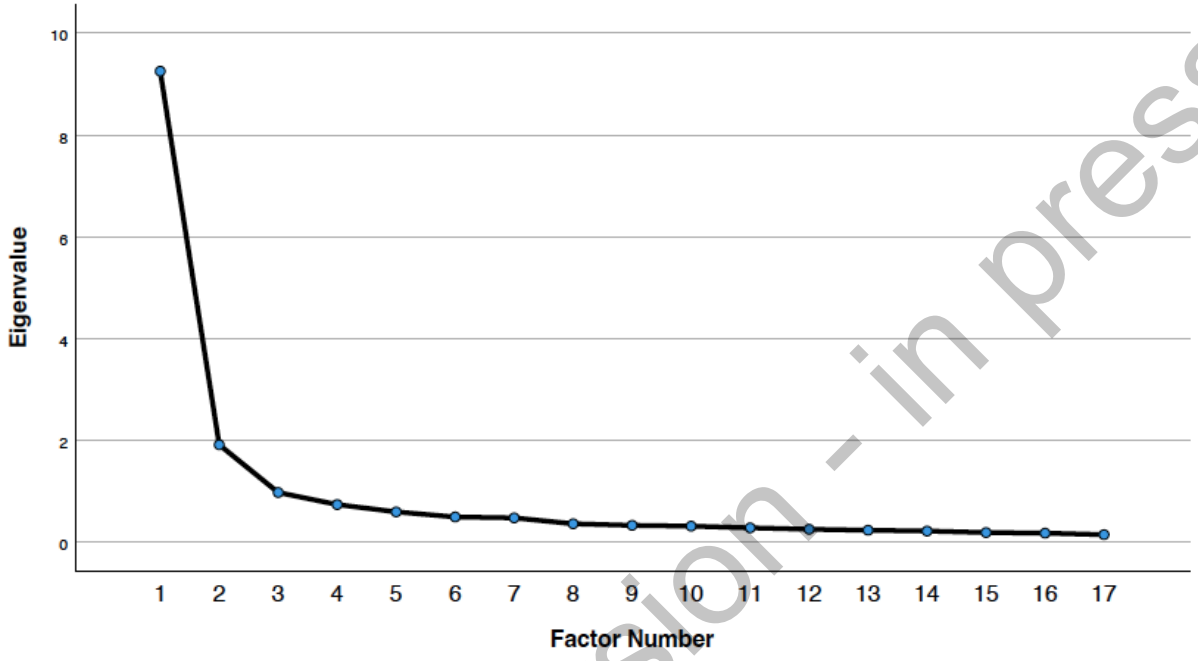

Figure S2A. Histogram showing distribution of frequency of responses in overall PrU

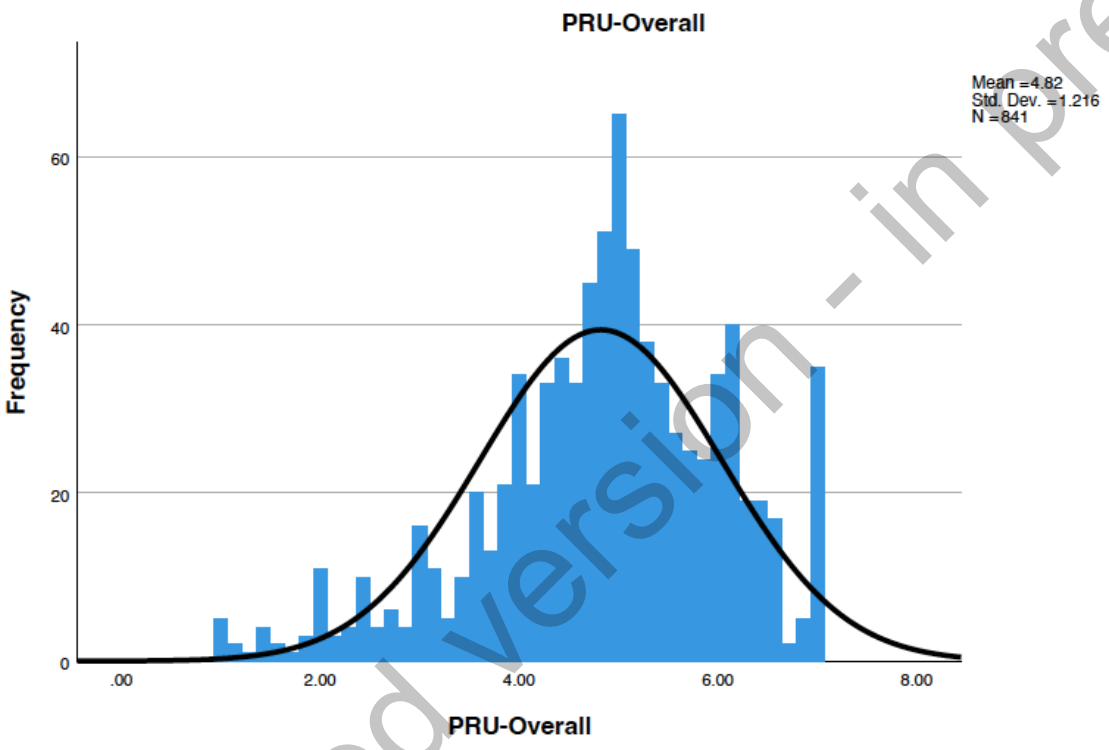

Figure S2B. Histogram showing distribution of frequency of responses in PrU self-knowledge subscale

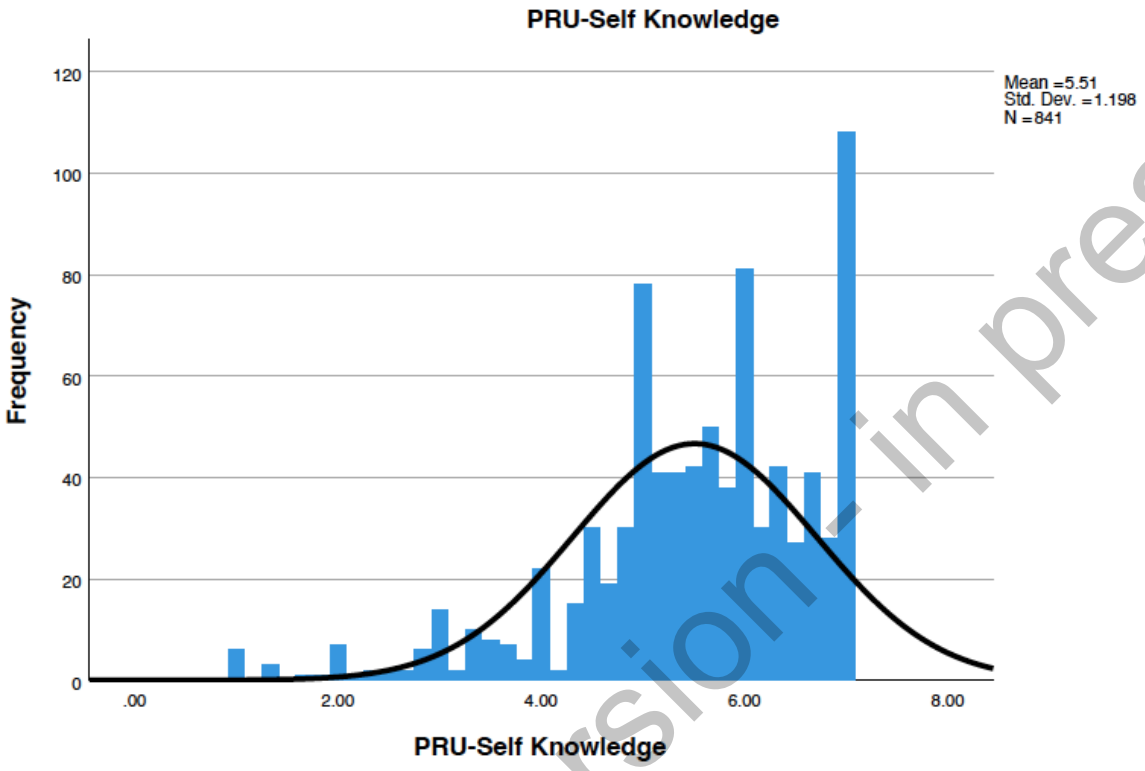

Figure S2C. Histogram showing distribution of frequency of responses in PrU reproductive planning subscale

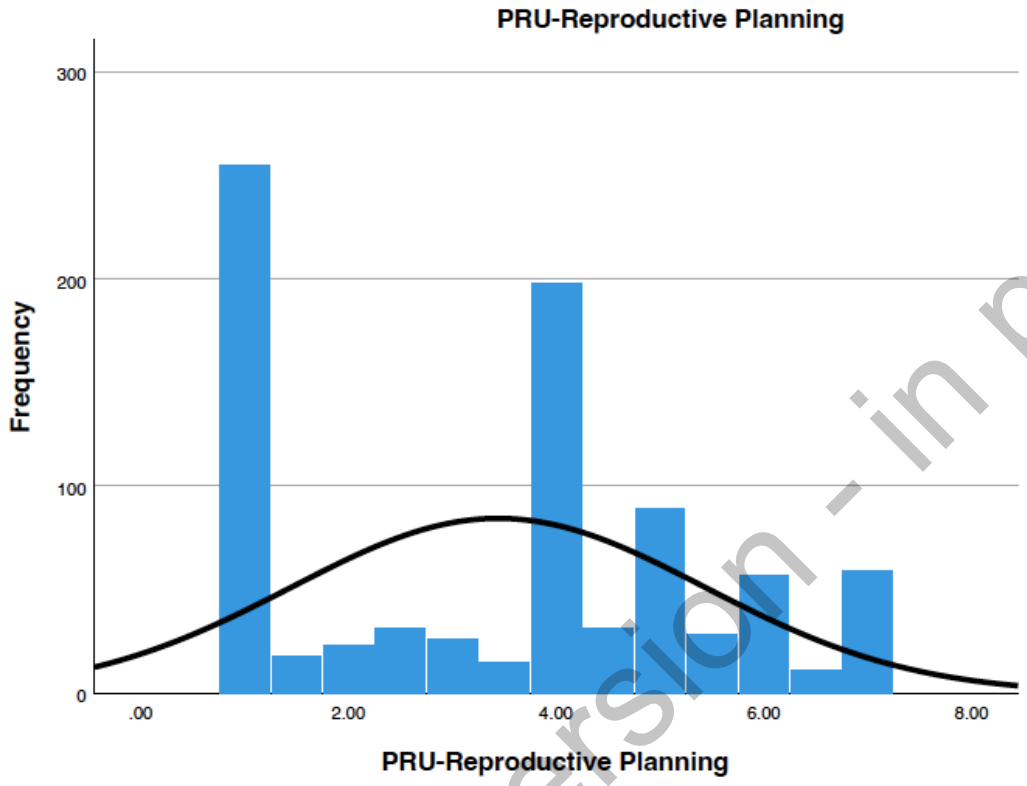

Figure S2D. Histogram showing distribution of frequency of responses in PrU practical benefits subscale

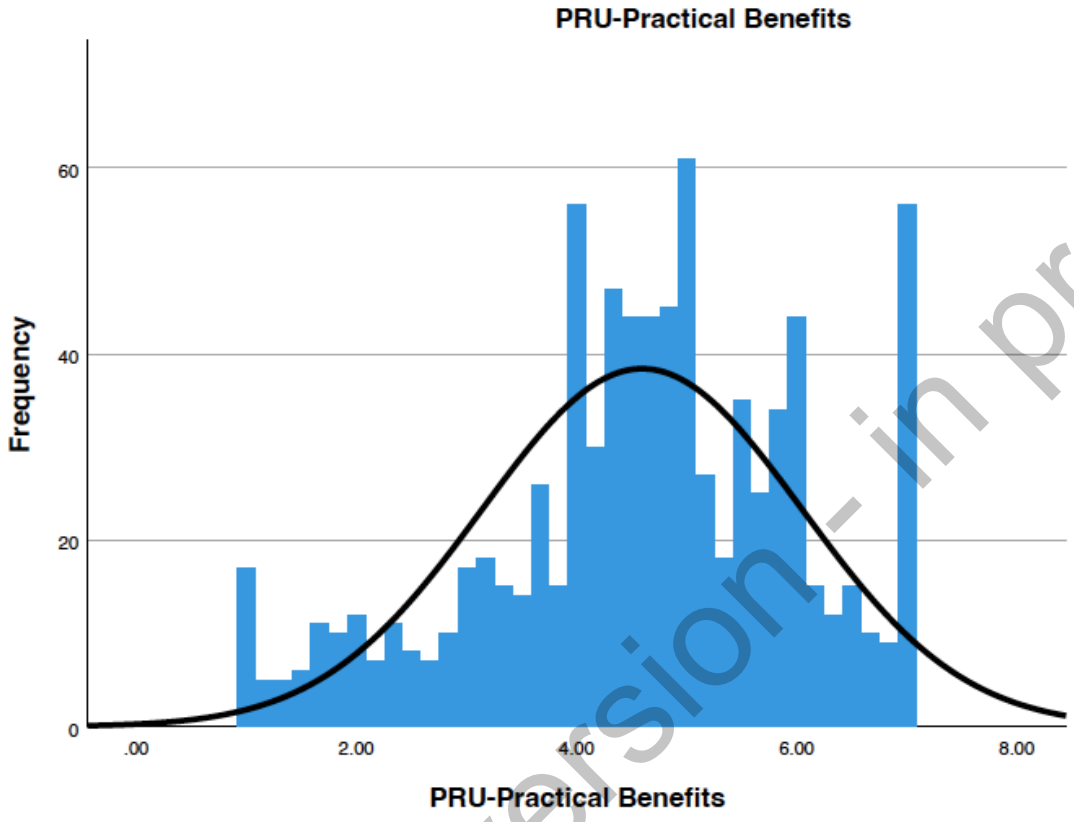

Table S1: Summary statistics and skewness of PrU items

		PRU1 Help with(my child's) life planning	PRU2 Inform my plans for (my child's) school or career	PRU3 Inform my (my child's) decisions about having children	PRU4 Use for testing a future pregnancy, if appropriate	PRU5 Help me or our family mentally prepare for the future	PRU6 Help to better understand my/my child's health	PRU7 Contribute to my /my child's self-knowledge	PRU8 Help me cope with my/my child's health risks	PRU9 Help me feel more in control of my/my child's health
N	Valid	841	841	841	841	841	841	841	841	841
	Missing	0	0	0	0	0	0	0	0	0
Mean		4.68	3.50	3.53	3.35	4.67	5.41	5.62	5.05	5.21
Median		5.00	4.00	4.00	4.00	5.00	6.00	6.00	5.00	5.00
Std. Deviation		1.736	1.969	2.103	2.083	1.811	1.465	1.352	1.596	1.565
Skewness		-.522	.066	.078	.170	-.615	-1.083	-1.228	-.836	-.928
Kurtosis		-.449	-1.141	-1.303	-1.292	-.462	1.003	1.582	.259	.448

		PRU10 Help me feel more in control of my/my child's life	PRU11 Simply to provide information	PRU12 Satisfy my curiosity (about my child)	PRU13 Help me/my child to use social programs, like resources and services	PRU14 Improve communication with my family members	PRU15 Feel good about helping the medical community	PRU16 Feel good about having information for family members	PRU17 Feel good about taking responsibility for my children's health
N	Valid	841	841	841	841	841	841	841	841
	Missing	0	0	0	0	0	0	0	0
Mean		4.90	5.53	5.61	3.94	4.34	5.46	5.41	4.77
Median		5.00	6.00	6.00	4.00	4.00	6.00	6.00	5.00
Std. Deviation		1.645	1.348	1.380	1.944	1.809	1.507	1.507	1.912
Skewness		-.707	-1.036	-1.263	-.187	-.432	-1.085	-1.158	-.678
Kurtosis		-.030	1.057	1.681	-1.015	-.629	.787	1.054	-.507

Table S2. Factor structure and factor loadings prior to item removal

	Factors		
PrU items	**1**	**2**	**3**
PRU11 Simply to provide information	.912		
PRU7 Contribute to my self-knowledge	.892		
PRU12 Satisfy my curiosity	.843		
PRU6 Help to better understand my health	.765		
PRU16 Feel good about having information for family members	.640		
PRU15 Feel good about helping the medical community	.620		
PRU9 Help me feel more in control of my health	.615		-.302
PRU17 Feel good about taking responsibility for my health	.322		
PRU3 Inform my decisions about having children		.937	
PRU4 Use for testing a future pregnancy, if appropriate		.882	
PRU14 Improve communication with my family members			-.806
PRU13 Help me to use social programs, like resources and services			-.726
PRU1 Help with life planning			-.533
PRU10 Help me feel more in control of my life	.411		-.500
PRU5 Help me or our family mentally prepare for the future			-.482
PRU8 Help me cope with my health risks	.432		-.480
PRU2 Inform my plans for school or career		.424	-.459