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QALYs in adults with cerebral palsy: Mapping from the San Martin Scale onto the EQ-5D-5L instrument

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Abstract

Background: Responses on health-related quality of life measured by disease-specific instruments can be mapped onto the EQ-5D-5L to estimate utility values for economic evaluation. San Martin´s Quality of Life Scale (St. MQoL-S) is a preferred measure to obtain health outcomes in adults with cerebral palsy. Nevertheless, it lacks a preference-based health utility score for estimating quality-adjusted life years (QALYs).

Objective: To develop algorithms for mapping from the St. MQoL-S to allow future prediction of the EQ-5D-5L, in adults with cerebral palsy, when utility data have not been collected.

Methods: Direct mapping models were developed using ordinary least squares, a generalized linear model, and Tobit regression analysis to estimate EQ-5D-5L utilities, with St. MQoL-S total and domain scores as explanatory variables, in a cross-sectional study of adults with cerebral palsy in Spain. Goodness-of-fit was assessed using mean absolute error (MAE) and root mean square error (RMSE). Repeated k-fold cross-validation was employed to select the optimal mapping model demonstrating superior predictive performance.

Results: The best-performing model for predicting EQ-5D-5L utilities, includes the St. MQoL-S total scores, age, gender, and types of cerebral palsy as explanatory variables in a stepwise ordinary least squares regression, making it the most robust model for use as a mapping algorithm with external data.

Conclusion: This is the first study to present mapping algorithms between the St. MQoL-S and EQ-5D-5L. The mapping functions preferred in this study seem adequate for estimating the utilities of the EQ-5D-5L for economic evaluation and to obtain QALYs in adults with cerebral palsy.

Keywords: St. MQoL-S, Cerebral palsy, EQ-5D-5L, QALYs, Economic Evaluation, Mapping, HRQoL.

INTRODUCTION

Cerebral palsy (CP) is a neurological condition that encompasses various permanent conditions of movement and/or posture, derived from brain injuries during development, in addition to the fact of suffering comorbidities such as epilepsy, intellectual disability, and sensory limitations [1]. Its incidence in Spain and Europe is 2-3 cases per 1,000 live births. Life expectancy is approximately 30-70 years [1,2]. It is a disease that has no cure, thus directly impacting on health-related quality of life (HRQoL) from pediatric age to old age.

The socio-economic repercussions of CP are diverse, especially in terms of specialized treatments such as rehabilitation and the high demand for meaningful care that these patients require to perform daily tasks [3]. The ongoing care that this condition requires can place considerable financial burdens on society as a whole [4,5]. Given the burden and economic impact of CP, it is crucial to improve our knowledge base to guide treatment, prevention, and the potential need for evidence to support health technology assessment and reimbursement decisions.

Currently, there is limited literature on health benefits based on the preferences of adult patients with CP [6], which is necessary to measure health effects, through the calculation QALYs for cost-effectiveness analyses. These types of economic evaluations usually take a social perspective, which means that all costs and effects are included in the analysis regardless of who bears them [7].

Several agencies recommend the use of QALYs in cost-effectiveness analyses, including; the Panel on Cost Effectiveness in Health and Medicine [8]. The EuroQol five-dimensional Questionnaire (EQ-5D-5L), is the preferred measure of the National Institute for Health and Care Excellence [9]. Besides, it is a generic short-term preference-based health outcomes instrument widely used around the world, which facilitates comparisons of health technologies across different diseases [7,10,11]. It has also proven to be a valid, concise, simple, and effective instrument for measuring HRQoL in people with CP [12,13]. However, most studies evaluating the quality of life in CP populations tend to favor disease-specific quality-of-life instruments [14], or clinical measures of performance status versus generic instruments such as the EQ-5D-5L [15,16].

When EQ-5D-5L data are not available, the National Institute for Health and Care Excellence allows utilities to be estimated by mapping other health-related measures of quality of life [9]. Several studies have been published related to the assignment of disease-specific quality of life instruments to the EQ-5D-5L [17,18] and guidelines for best practices have been developed [19,20]. Mapping a non-preference-based measure to the EQ-5D-5L can be performed by predicting the health utility values of the EQ-5D-5L (direct mapping) or each of the five domain responses (indirect mapping). However, there is limited use of either approach in the context of neurological diseases [13,21,22].

The San Martin Scale (St.MQoL-S) is a recommended and frequently used instrument in institutions specialized in the care of adults with CP in different countries [23–26], as an outcome measure and proxy for HRQoL, and to determine the effectiveness of treatments and interventions [24,25]. The most important reason for choosing this type of specific HRQoL instruments is that they are already applied in the clinical practice of institutions that are dedicated to the care of these adults and, therefore, the staff is used to working with them. In addition, they tend to be more sensitive to differences or changes in the health status of individuals with a specific disease [13,14].

The appropriateness of existing generic HRQoL measures for application in adults with CP, has received limited research attention [6,27]. In addition, a mapping algorithm specific to adults with CP using a generic instrument for this population, has not yet been developed. Given that some relevant items and/or domains overlap between the St. MQoL-S and the EQ-5D-5L (see TableS1 in the supplementary material), it is plausible that there is a significant relationship between these two instruments. Consequently, we considered developing a mapping algorithm that would allow the calculation of QALYs when investigators wish to explore the cost- effectiveness of an intervention.

This study aimed to develop a mapping algorithm from the St. MQoL-S to allow future prediction of the usefulness of the EQ-5D-5L, in adult patient populations with CP, when utility data have not been collected.

METHODS

Study Sample

Data were obtained from the Study of Adults Cerebral Palsy in Navarre, Spain (EPCANA). This cross-sectional study of quality of life and clinical outcomes was carried out at the Spanish Association for the Care of People with Cerebral Palsy (Aspace), within the framework of a comprehensive assessment of HRQoL to obtain QALYs, through a specific instrument (St.MQoL-S) [25,28] and a generic instrument (EQ-5D-5L) [11]. Participants completed a series of outcome measures and provided demographic and clinical information. For the analysis, we used responses from a representative sample of patients recruited by Aspace's clinical and research teams up to July 2023. Cross-sectional data from 72 participants who completed both the St. MQoL-S and the EQ-5D-5L at the same time, were used to develop and identify the best-performing mapping algorithm.

The study used the functional classification of the recognized level of dependence, which is composed of three levels, level I represents the mildest level of dependency and level III represents the most severe. This classification is included in the battery of data collected by the San Martin Scale to identify the person being evaluated. On the other hand, the classification of the types of CP was also used, based on neurological findings: spastic, dyskinetic, ataxic, and unclassifiable.

The EPCANA received ethical approval for its realization in Aspace Navarra and was registered with the Public University of Navarra (no. 026/23).

Measures

The EQ-5D-5L was included in the EPCANA study to estimate health utility values. It is an objective measure by which patients describe their state of health [11]. It covers the health domains of mobility, self-care, usual activities, pain/discomfort, and anxiety/depression. Respondents can indicate one of five levels of severity: no problems, mild problems, moderate problems, severe problems, and extreme problems in the corresponding health dimension [29]. A preference-based single index score can be generated with any combination of responses, anchored at 0 to represent death and 1 to represent full health, and based on a Spanish rate [30]. These health utility values have been developed using the general public's responses to a standard gambling survey [30].

The St. MQoL-S evaluates a comprehensive, functional, and specific approach to quality of life [25]. It consists of a validated 95-item questionnaire specific to adults with significant disabilities, including CP. The responder is asked to answer the questions with a ‘never’, ‘sometimes’, ‘often’ or ‘always’. Responses to St. MQoL-S are often used to derive a single HRQoL index value and this value is reported in clinical studies. The St. MQoL-S measures HRQoL over a range of values from 52 to 132 points. A higher score represents higher HRQoL in the St. MQoL-S index or, in other words, St. MQoL-S total score. This scale measures HRQoL in 8 dimensions (Figure 1)

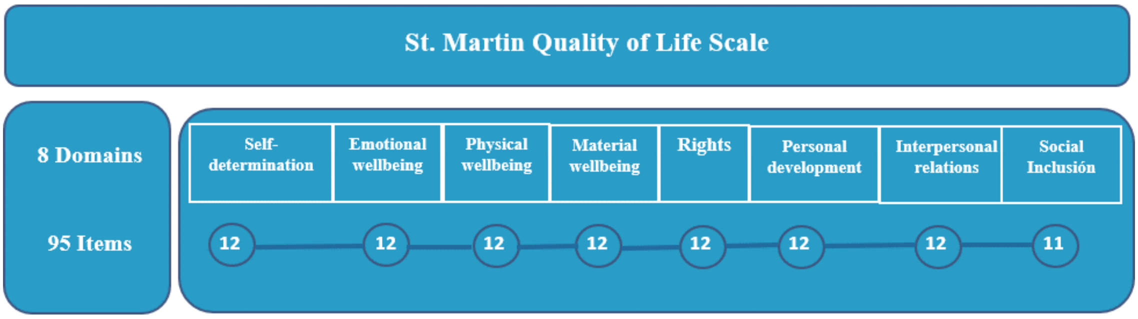


Figure 1: Structure of the San Martin Scale, showing the breakdown into eight domains and ninety-five items. The items are related in each dimension. All dimensions have 12 items, except for the social inclusion domain, which has 11 items. To see the full scale, (see supplementary material, at the bottom of Table S1). Own elaboration, based on the Scale Manual [31].

It is a good measurement instrument in terms of reliability and validity that is mainly used in institutions dedicated to the specialized care of adults with CP [32]. Several studies have attestedto its sensitivity, and that it is suitable for direct and proxy assessment in disabilities such as CP [23,24,26]. For this reason, it is recommended not only for scientific research purposes but also inclinical practice [31].

Statistical analysis

Mapping from St. MQoL-S to the EQ-5D-5L

To develop a crosswalk between the St. MQoL-S and the EQ-5D-5L in the EPCANA study, Anova F-test and stepwise regression were used to examine alternative models and predictor variables, ultimately selecting an optimal mapping model. Two core models, based on direct additive mapping, were used to regress the utility score of the EQ-5D-5L on the general or domain scores of the St. MQoL-S [9,18]. An item response level (indirect) mapping approach was not chosen because it requires a large sample size with an adequate number of responses for each response level, which was not available in this study [9,21,33]. The first core model is based on the St. MQoL-S total score, whereas the second core model is built upon the St. MQoL-S domain total scores. In both cases, a combination of independent socio-demographic (age andgender) and clinical variables (type of CP and level of dependence) are considered to predict the utilities of the EQ-5D-5L[9,17,20,21,34] (based on the Spanish rate [30]).

In all mapping analyses, we investigate which explanatory variables serve as potential predictors for the utility score of the EQ-5D-5L. We conclude that a combination of age, gender, type of CP (CPT), and the St. MQoL-S total score represents the optimal choice for developing the mapping algorithm in the first core model. Concerning the second core model, a stepwise selection was carried out to identify the significant domains for a more efficient mapping. The domains of self-determination, physical well-being, material well-being, rights and personal development were selected as predictor variables in the model, together with the same socio-demographic and clinical variables included in the first core model.

The regression equations of the two core models are represented as follows:

$$\boldsymbol{EQ-5D\ Index} = \alpha + \beta_1 * St.MQoLS_{Total\ Score} + \beta_2 * Age + \beta_3 * Sex + \beta_4 * CPT \quad (Model\ 1)$$

$$\boldsymbol{EQ-5D\ Index} = \alpha + \sum_{j=1}^{5} \beta_j * St.MQoLS_{domain_j} + \beta_6 * Age + \beta_7 * Sex + \beta_8 * CPT \quad (Model\ 2)$$

where the EQ-5D Index is the EQ-5D-5L utility scores and $St.MQoLS_{domain_j}$ represents the St. MQoL-S domains selected according to statistical significance (p <0.05) using stepwise regression.

Three different regression methods were considered for each model candidate of direct mapping. Firstly, we employed *Ordinary Least Squares* (OLS) to fit a linear model over the response variable, a method widely used in previous studies with acceptable performance [9,17,18]. Due to the non-normal distribution of the data of the EQ-5D-5L, a *Generalized Linear Model* (GLM) with a Gaussian family and a logit link function was chosen as a second model candidate. Finally, we also considered a *Tobit regression model* [35] to address censored data at the upper limit of 1 in the response variable.

Data management was carried out using Microsoft Excel, and R statistical software version 4.3.2 [36] was used for statistical analysis.

Evaluating model performance

Following the mapping guide [9,19], we assess the goodness-of-fit of each model candidate by evaluating the predicted values using mean absolute error (MAE) and root mean squared error (RMSE).

Since our objective is to develop a mapping algorithm that can be generalized to other individuals and populations, we also conduct a cross-validation approach to validate the predictive performance of the models [37]. Given the absence of a dataset for external validation, internal validation was performed by considering a 5-fold cross-validation scheme. We first divide the sample into five random groups. Four groups (80% of the sample) were assigned as the "estimation sample", used for estimating the models regression coefficients, while the remaining group (20% of the sample) form the "validation sample", which was used to evaluate the predicted EQ-5D-5L scores for the various model candidates. This procedure is repeated so that each of the five random groups served as a validation sample. To obtain more reliable and robust estimates of prediction errors, assessed by MAE and RMSE values, a repeated 5 -fold cross-validation method was implemented.

RESULTS

Table 1 presents the characteristics of the 72 adults constituting our sample. The EQ-5D-5L utility score was not normally distributed ($p < 0.05$). The normality test was not rejected for the total score of the St. MQoL-S; the distribution was slightly skewed to the left (Figure S1). The mean (SD) EQ-5D-5L utility was 0.36 (0.23) and the mean total scores for the St. MQoL-S was 104.40 (10.40). Approximately more than half (61.11%) of the adults were male. The mean age was 39.25 years (SD 15.77 years).

Table 1. Characteristics of the sample (n=72)

Quantitative Variable	**Mean (SD)**
St. MQoL-S total score	104.40 (10.42)
St. MQoL-S Self-determination domain	12.82 (2.30)
St. MQoL-S Emotional Well-being domain	11.07 (2.17)
St. MQoL-S Physical Well-being domain	10.85 (2.09)
St. MQoL-S Material Well-being domain	10.65 (2.14)
St. MQoL-S Rights domain	11.49 (2.08)
St. MQoL-S Personal Development domain	10.53 (2.70)
St. MQoL-S Interpersonal relationships domain	9.44 (2.48)
St. MQoL-S Social Inclusion domain	9.26 (2.84)
EQ-5D Index, Mean (SD)	0.36 (0.23)
Age in years, Mean (SD)	39.25 (15.77)
Qualitative variables	**N (%)**
Gender	
Female	28 (38.89)
Male	44 (61.11)
Patients living outside the city	36 (50.00)
Patients living outside the city	36 (50.00)
Cerebral palsy type	
Spastic CP	50 (69.44)
Dyskinetic CP	4 (5.56)
Ataxic CP	3 (4.17)
Unclassified CP	15 (20.83)
Functional classification of the recognized level of dependency	
Mild	10 (13.89)
Moderate	16 (22.22)
Severe	46 (63.89)

CP: Cerebral Palsy; EQ-5D index: EQ-5D-5L utility scores; N: Sample size; SD: standard deviation; St. MQoL-S: St Martin's Quality of Life Scale.

Assessing the Correlation Between HRQoL Measures

A strong positive linear correlation ($r = 0.77$, $p < 0.05$) was found between EQ-5D-5L utility scores and St. MQoL-S total scores (Figure 2). At the domain level, a high correlation was found between the EQ-5D-5L and the St. MQoL-S domains ($p < 0.05$). Table S2 (supplementary material) presents the correlation coefficients for each domain. Figure S2 (supplementary material) presents the correlations between each St. MQoL-S domain and the EQ-5D-5L.

Mapping model performance

Table 2 presents the goodness-of-fit results of the different models, employing the entirety of the data to estimate the regression coefficients of the explanatory variables. The predicted average utility scores of the EQ-5D-5L are consistently close across the model candidates. Almost all models overestimated the lower bound of the EQ-5D-5L utility scores, while simultaneously underestimating its upper limit.

Figure 2: Scatter plot between the EQ-5D utility scores and St. MQoL-S total scores

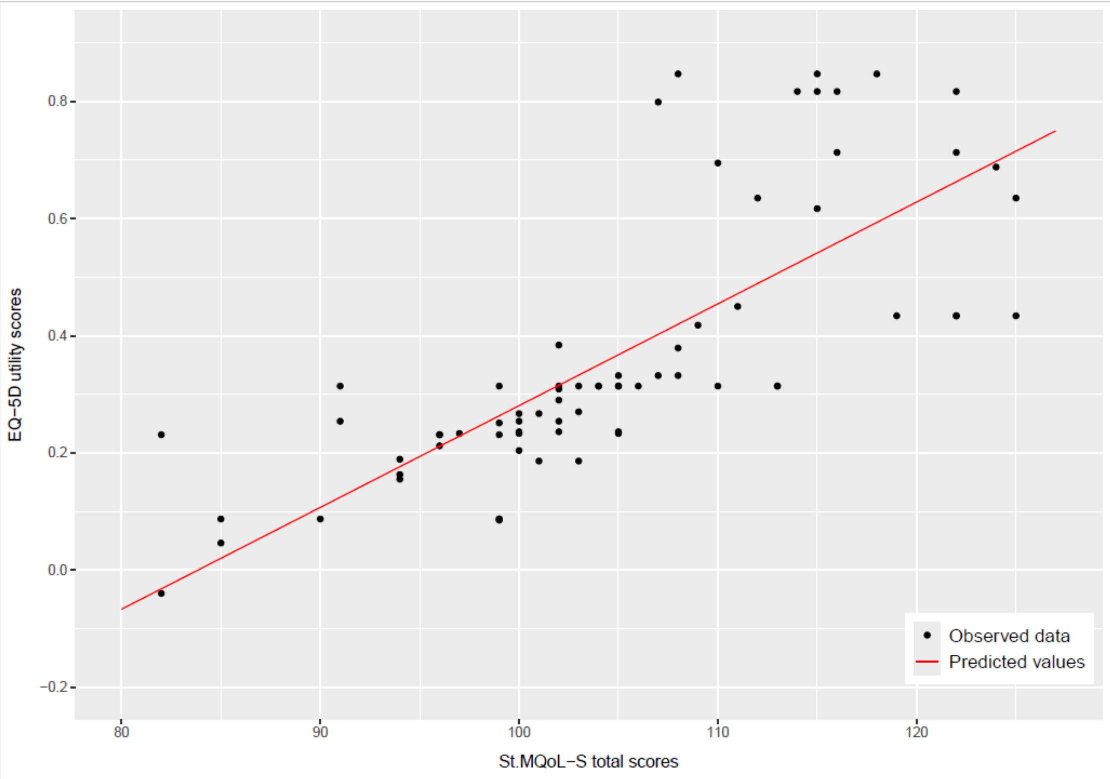


Table 2. Goodness-of-fit results of model candidates

Model specification (n=72)	Differences between predicted and observed mean	Mean predicted	Minimum predicted	Maximum predicted	MAE	RMSE
EQ-5D-5L observed utility	NA	0,3571	-0,202	0,8470	NA	NA
Method 1: Ordinary Least Squares Estimator						
Model 1	0,0000	0,3571	-0,0569	0,7258	0,1000	0,1367
Model 2	0,0000	0,3571	-0,1649	0,7221	0,0990	0,1326
Method 2: Generalized Linear Model: Gaussian family logit link						
Model 1	0,0043	0,3614	0,0650	0,7501	0,0978	0,1319
Model 2	0,0019	0,3590	0,0359	0,7446	**0,0971**	**0,1281**
Method 3: Tobit Estimator						
Model 1	0,0000	0,3571	-0,0569	0,7258	0,1000	0,1367
Model 2	0,0000	0,3571	-0,1649	0,7221	0,0990	0,1326

EQ-5D-5L: five-level EuroQol five-dimensional questionnaire; MAE: mean absolute error; RMSE: Root mean squared error; NA: not applicable.

In terms of goodness-of-fit, GLM model 2 showed the best predictive ability, with the lowest MAE (0.0971) and RMSE (0.1281), followed by GLM model 1 (MAE=0.0978, RMSE: 0.1319). Additionally, the GLM estimator of model 1 closely predicted the maximum EQ-5D-5L score, while the OLS estimator of model 2, as well as the Tobit estimator of model 2, closely predicted the minimum value.

Regression coefficient estimates for the different model candidates are presented in Table S3 (supplementary information online). In model 1, the St. MQoL-S total score was a significant predictor variable ($p<0.05$) in all three regression methods. In model 2, the self-determination domain on the St. MQoL-S was consistently significant in all three regression methods. The physical well-being domain and the rights domain were significant in the OLS and Tobit estimator, while material well-being was significant in the GLM and Tobit estimator. The personal development domain was significant only in the Tobit estimator. Age was significant in the GLM and Tobit estimator ($p<0.1$) and type of CP was also a significant predictor ($p<0.05$).

It should be recalled that all the variables mentioned in Table S3, were significant with the Anova F-test when selecting the explanatory variables, most appropriate for predicting the EQ- 5D-5L utility score. This was reflected in the variables that were retained in the two core models described above in the statistical analysis, in order to obtain the best possible mapping algorithm. We clarify that sex was not significant, but we decided to include this variable because of its ability to improve the mapping algorithm and its importance as a covariate in other populations.

Validation of mapping algorithms

Although GLM model 2 was initially identified as the best model to predict EQ-5D-5L utilities from the St. MQoL-S scores and the explanatory variables within our observed data, a cross-validation approach has been conducted to ensure generalization of this finding for out-of-sample individuals. Table 3 displays the result obtained under a repeated 5-fold cross-validation scheme.

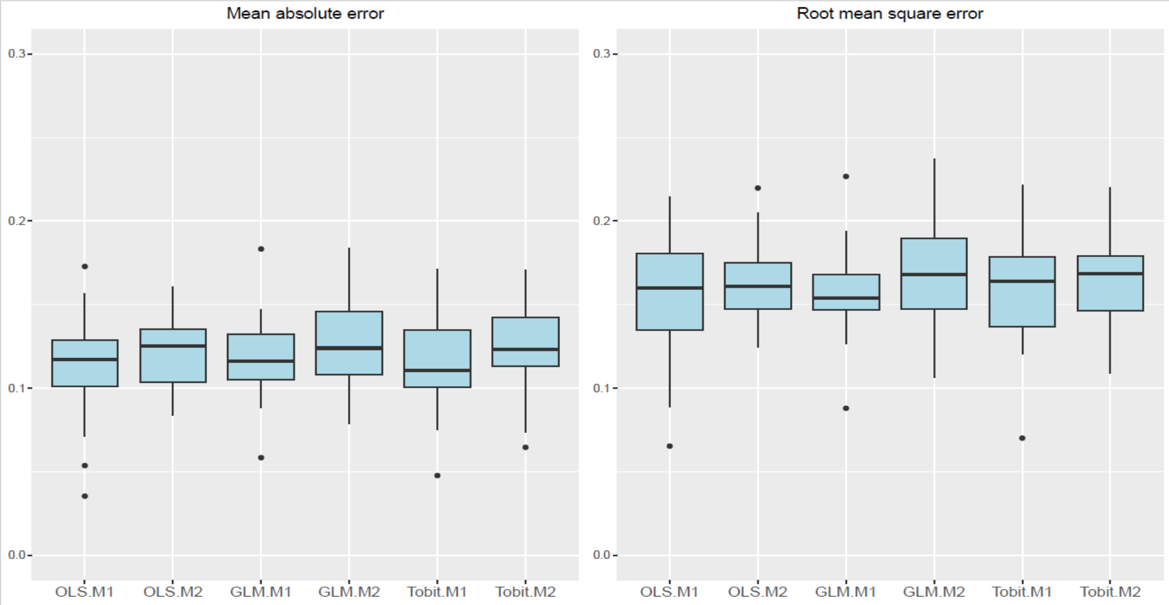
Table 3. Goodness of fit results using a repeated 5-fold cross-validation scheme.

Model specification (repeated 5-fold CV)	Differences between predicted and observed mean	Mean predicted	Minimum predicted	Maximum predicted	MAE	RMSE
EQ-5D-5L observed utility	NA	0,3571	-0,202	0,8470	NA	NA
Method 1: Ordinary Least Squares Estimator						
Model 1	0,0019	0,3590	-0,0986	0,8703	**0,1131**	**0,1545**
Model 2	0,0016	0,3587	-0,1918	0,8773	0,1245	0,1649
Method 2: Generalized Linear Model: Gaussian family logit link						
Model 1	0,0030	0,3601	0,0541	0,8140	0,1179	0,1595
Model 2	0,0060	0,3631	0,0310	0,8776	0,1282	0,1682
Method 3: Tobit Estimator						
Model 1	0,0002	0,3573	-0,0901	0,8866	0,1140	0,1562
Model 2	0,0004	0,3575	-0,1904	0,9148	0,1230	0,1651

EQ-5D-5L: five-level EuroQol five-dimensional questionnaire; MAE: mean absolute error; RMSE: Root mean squared error; NA: not applicable.

According to this analysis, the OLS model 1 exhibits the best predictive performance with lowest MAE (0.1131) and RMSE (0.1545) values. This suggests its preference as the mapping algorithm of choice for applications in other populations that have not collected utility data. However, overall differences between OLS, GLM, and Tobit models were small (Figure 3). The scatter plots for the observed and predicted EQ-5D utility scores of all mapping models are shown in Figures S3 and S4 (supplementary material).

Figure 3: Box plots of estimated prediction error measures among different cross-validation samples.



The OLS model 1 can be presented as follows:

$$\boldsymbol{EQ-5D\ index} = -1.380 + 0.016 * St.MQoLS_{Total\ Score} + 0.002 * Age + 0.002 * I(Sex = Male) + 0.078 * I(CPT = Dyskinetic) + 0.016 * I(CPT = Ataxic) + 0.084 * I(CPT = Unclassified)$$

where the symbol I (·) denotes an indicator function.

After obtaining the mapping algorithm that best fits the data, the QALY per year for an adult person with CP can be obtained. For example, a person who has a St. MQoL-S total score of 106, is 30 years old, is female (0), and has a CP of the spastic type, her score would be:

$$\boldsymbol{QALY}_{\boldsymbol{Per\ year}} = -1.380 + 0.016 * (106) + 0.002 * 30 = 0.3357$$

Methodological reproducibility and replicability

The original data and R code to fully reproduce and replicate the analyses presented in this study are available as supplementary material (see Data_and_Rcode.zip file). Additionally, a README file has been included providing a comprehensive description of the data and detailed R scripts to obtaining the best algorithm used for mapping St. MQoL-S to EQ-5D-5L. This allows for a more thorough understanding of the methodology employed in the current analysis. Referring to this additional resource can enhance the appreciation and critical evaluation of the results obtained, as well as provide greater transparency in the methodological process used in the research. For examples illustrating the usage of the mapping algorithm, please refer to the examples section within the README file.

DISCUSSION

Our study provides evidence that the St. MQoL-S could conceptually serve as a promising candidate for mapping to the EQ-5D-5L in adult patients with CP. This could be partly explained by the fact that our mapping results fall within the MAE and RMSE ranges of other mapping studies [17] and allowed us to affirm the viability of mapping from the St. MQoL-S to the EQ-5D-5L. Our mapping models showed significantly better predictive outcomes when utilizing the St. MQoL-S total score/index as an explanatory variable instead of scores from the most significant dimensions (Self-determination, Physical wellbeing, Material well-being, Rights and Personal development) when estimating EQ-5D-5L utilities. This enhancement can be attributed to the robust correlation observed between the total scores of the two HRQoL scales and the specific characteristics of the study population [21,33].

Consistent with many previous mapping studies, our validation analysis indicates that Ordinary Least Squares (OLS) regressions demonstrated the highest predictive performance, showing a slightly improvement upon the results of Tobit regressions for direct mapping [9,17]. Consequently, we consider the Tobit model could also serve as a potential mapping algorithm, effectively constraining predicted values of the response variable, and thereby proving particularly valuable when analysing censored data.

Our preferred OLS model 1, which utilizes the total score of the St. MQoL-S and other socio-demographic and clinical explanatory variables such as age, gender and CP type, exhibited MAE and RMSE values comparable to those reported in previous neurological statistical mapping works [21,33], as well as in other mapping studies overall [34].

The discrepancy between the model exhibiting the highest goodness-of-fit and the model selected for superior predictive performance underscores the critical importance of conducting standard cross-validation techniques for mapping algorithms [9,20].

The mapping methodology offers a valuable tool for integrating, comparing and synthesizing data from different sources and health outcome measures to obtain QALYs in cost effectiveness analyses [38]. This technique facilitates the extrapolation of data when health utility data are not collected directly, thus allowing a more complete estimation of the impact of health interventions related to the HRQoL [18,19,34] of patients with pathologies as specific as cerebral palsy [21]. In addition, mapping enables direct comparison of the relative effectiveness and efficiency of different intervention strategies, which is essential for informed decision making in health policy and resource allocation in public health [9,20].

This study constitutes a significant addition to the economic evaluation literature concerning population with cerebral palsy. It presents results for additive direct mapping algorithms utilizing various model structures, facilitating the predicting health utility values. Previous mapping studies in CP populations have been limited in number [18,21]. Our study, to the best ofour knowledge, is the first to conduct such analysis within an adult population with cerebral palsy, providing valuable evidence for the development of economic evaluations when EQ-5D- 5L data have not been directly collected. An important strength of the analysis lies in the integrity of the returned questionnaires, with no evidence of missing data, as patients were always accompanied when responding to questionnaires to clarify doubts, enable simultaneous completion of both questionnaires, and avoid unanswered questions.

The main limitation of our study could be the small number of patients analysed, which would have rendered our findings more robust with a larger sample. As a cross-sectional study, EPCANA provided the opportunity to measure multiple variables to increase study power, but with only 72 participants, the sample size was deemed limited for such analysis. Nevertheless, the recommended mapping algorithm can be useful for conducting cost-utility analyses in similar populations. The GLM models in this study showed slightly poorer results, as they do not allow for predicting negative EQ-5D-5L utility values for CP patients and had a higher error rate for low utility scores. This is concerning because CP is associated with relatively low utility values reflecting very poor HRQoL, although in our data only a few patients reported negative utilities (2.7%). The mapping algorithms presented in this study were validated from a sample of data arising from the same study. While this is a common technique in the literature [21,33,34], external validation would have been preferable in the context of a broader generalization assessment. Lastly, it should be noted that directly collected EQ-5D-5L utility values always supersede predicted values based on mapping algorithms.

Further research is needed to better capture health state utilities in adults with CP, such as developing specific instruments to measure better HRQoL CP aging stages and validating the mapping algorithm using a large sample with an external dataset.

CONCLUSIONS

Several studies focusing on quality of life in populations affected by CP have dispensed with the use of preference-based utility measures, despite their increasing need to inform decisions in economic evaluations needed to assess the quality of treatments in CP. The algorithms presented in this study offer an alternative for estimating the utility of the EQ-5D-5L in scenarios where utility information has not been directly collected from CP patients. Our research has demonstrated the feasibility of predicting, with reasonable accuracy (based on MAE and RMSE results reported in other mapping studies), the utility values of the EQ-5D-5L derived from the St. MQoL-S. These findings should help the economic evaluations of interventions for adults with CP by providing evidence linking the St. MQoL-S, an outcome measure of interventions aimed at the specialized care of adults with CP used in different countries, to a generic preference-based tool that is widely used around the world, the EQ-5D-5L.

Declarations

Ethical Approval

The Study of Adults Cerebral Palsy in Navarre, Spain (EPCANA) received ethical approval for its realization in Aspace Navarra and was registered with the Public University of Navarra (no. 026/23). All participants involved in the study signed an informed consent form to participate and publish their data.

Data availability

The data and codes that were used to obtain the results of this manuscript are available in the supplementary material attached to this manuscript, inside a compressed zip file (Data_and_Rcodef) containing a file with README link, where you can see the data and how to replicate the codes.

Conflict of interest

The authors have no conflict of interest

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Table S1: Comparison between the domains of the scales: St. MQoL-S and EQ-5D-5L

EQ-5D-5L domains	San Martin Quality of Life Scale (St. MQoL-S)	
	Items	**Domains**
Mobility	Specific measures are taken in relation to their mobility in order to encourage their independence	Physical well-being
	Moves through different spaces (i.e., indoors and outdoors)	
	Specific measures are taken to prevent or treat problems arising from physical disabilities (e.g., spasticity, stiffness, deformity, etc.)	
	Specific measures are taken in relation to their mobility in order to encourage their independence	
Selfcare	Specific measures are taken by the institution to avoid risks such as blows, falls and escapes.	Material well-being
	Specific measures are taken to adapt the living environment to the person's abilities and limitations (i.e. sensory, cognitive, behavioural, physical).	
	Chooses how to spend leisure time.	Self-determination
	Chooses the meal or part of the meal when there is variety in 1st, 2nd and dessert.	
	Has a daily schedule of activities appropriate to his/her preferences	
	They have a personal record of what they like, what calms them, what they cannot tolerate and how they may react, which all staff are aware of and must comply with.	Emotional well-being
	Appropriate love, affection and physical contact are provided when needed.	
	Participates in inclusive activities appropriate to their physical and mental condition	Social Inclusion
	Participates in inclusive activities that interest him/her	
Usual activities	You choose how you spend your free time	Self-determination
	Has opportunities to refuse to do activities that are irrelevant to his/her health	

	chooses the meal or part of the meal when there is variety in 1st, 2nd and dessert	
	Decorates the room to his liking	
	Has a daily schedule of activities appropriate to his/her preferences	
	Performs physical activities and exercises appropriate to his or her characteristics and needs.	Physical well-being
	Has adequate hygiene (e.g., teeth, hair, nails, body) and personal image (e.g., age-appropriate clothing, occasion-appropriate clothing, etc.). (e.g., clothing appropriate to age, occasion, etc.	
	You have a programme of activities with things that you enjoy and contribute to your personal enrichment.	Personal Development/Rights
	The activities you do allow you to learn new skills.	
	You are taught things that interest you	
	You learn things that make you more independent	
	At the centre you have opportunities to show your skills	
	Specific measures are taken to teach him new skills	
	You gain new skills or experiences by participating in activities.	
	Participates in activities outside the centre with people outside his/her support context.	Social Inclusion
	The activities in which he/she participates consider the leisure and cultural facilities in the area.	
	Specific measures are taken to offer as wide a variety of activities as possible (e.g., new activities based on people's preferences).	
	Participates in social activities outside the place where he/she receives services or supports	
Pain/Discomfort	Specific measures are taken to prevent or treat pain	Physical well-being
Anxiety/Depression	Supporters are aware of your individual expressions of distress.	Emotional well-being

For more detailed information on the San Martín Quality of Life Scale and EQ-5D-5L instrument please consult the following link:

https://sid-inico.usal.es/idocs/F8/FDO26729/San_Martin_Scale_English_(Verdugo_Gomez_et_al_2014).pdf

https://euroqol.org/information-and-support/euroqol-instruments/eq-5d-5l/

Table S2: Spearman correlation coefficients between EQ-5D utility score and domains of St. MQoL-S total scores

	EQ-5D utility scores	St. MQoL-S Self-determination domain	St. MQoL-S Emotional Well-being domain	St. MQoL-S Physical Well-being domain	St. MQoL-S Material Well-being domain	St. MQoL-S Rights domain	St. MQoL-S Personal Developmental domain	St. MQoL-S Interpersonal Relationships domain	St. MQoL-S Social Inclusion domain
St. MQoL-S Self-determination domain	0.68*								
St. MQoL-S Emotional Well-being domain	0.55*	0.47*							
St. MQoL-S Physical Well-being domain	0.65*	0.35*	0.43*						
St. MQoL-S Material Well-being domain	0.60*	0.45*	0.48*	0.56*					
St. MQoL-S Rights domain	0.59*	0.26*	0.53*	0.61*	0.55*				
St. MQoL-S Personal Developmental domain	0.66*	0.50*	0.61*	0.47*	0.49*	0.56*			
St. MQoL-S Interpersonal Relationships domain	0.67*	0.43*	0.58*	0.65*	0.55*	0.59*	0.70*		
St. MQoL-S Social Inclusion domain	0.67*	0.49*	0.15	0.43*	0.24*	0.18	0.37*	0.35*	

*Note * p-value <0.05*

Table S3: Estimated regression coefficients of model candidates

Independent variables	Ordinary Least Squares estimator	Generalized Linear Model	Tobit Model
Panel A - Model 1			
St. MQoL-S total scores	0.016 (0.002) ****	0.075 (0.010) ****	0.016 (0.002) ****
Age (year)	0.002 (0.001)	0.010 (0.006) *	0.002 (0.001) *
Sex	0.002 (0.035)	0.007 (0.174)	0.002 (0.034)
CP Dyskinetic	0.078 (0.078)	0.293 (0.328)	0.078 (0.074)
CP Ataxic	0.166 (0.088) *	0.604 (0.378)	0.016 (0.084) **
CP Unclassified	0.084 (0.043) *	0.390 (0.200) *	0.084 (0.041) **
Constant	-1.380 (0.178) ****	-9.041 (1.031) ****	-1.380 (0.169) ****
Panel B - Model 2			
St. MQoL-S Self-determination domain	0.025 (0.010) **	0.131 (0.048) ***	0.025 (0.009) ***
St. MQoL-S Physical Well-Being domain	0.020 (0.011) *	0.091 (0.055)	0.020 (0.010) *
St. MQoL-S Material Well-Being domain	0.018 (0.012)	0.104 (0.054) *	0.018 (0.011) *
St. MQoL-S Rights domain	0.022 (0.013) *	0.107 (0.070)	0.022 (0.012) *
St. MQoL-S Personal Development domain	0.014 (0.009)	0.058 (0.041)	0.014 (0.008) *
Age (year)	0.001 (0.001)	0.006 (0.006)	0.001 (0.001)
Sex	-0.005 (0.037)	-0.040 (0.184)	-0.005 (0.034)
CP Dyskinetic	0.100 (0.081)	0.422 (0.345)	0.100 (0.074)
CP Ataxic	0.188 (0.089) **	0.712 (0.389) *	0.188 (0.082) **
CP Unclassified	0.070 (0.044)	0.302 (0.206)	0.070 (0.041)
Constant	-0.838 (0.126) ****	-6.651 (0.799) ****	-0.838 (0.116) ****

CP Spastic=1, CP Dyskinetic=2, CP Ataxic=3, CP Unclassified=4
Model 1 uses total St. MQoL-S scores to predict EQ5D utility scores;
Model 2 uses St. MQoL-S domain scores to predict EQ5D utility scores
Sex= Female=0, Male=1
**** Significant at p<0.001; ***Significant values at p<0.01; **Significant values at p<0.05; * Significant values at p<0.1
Standard errors in parentheses

Figure S1: Distribution of EuroQOL (EQ-5D-5L) utility scores and San Martin Quality of life scale (St. MQoL-S) total scores.

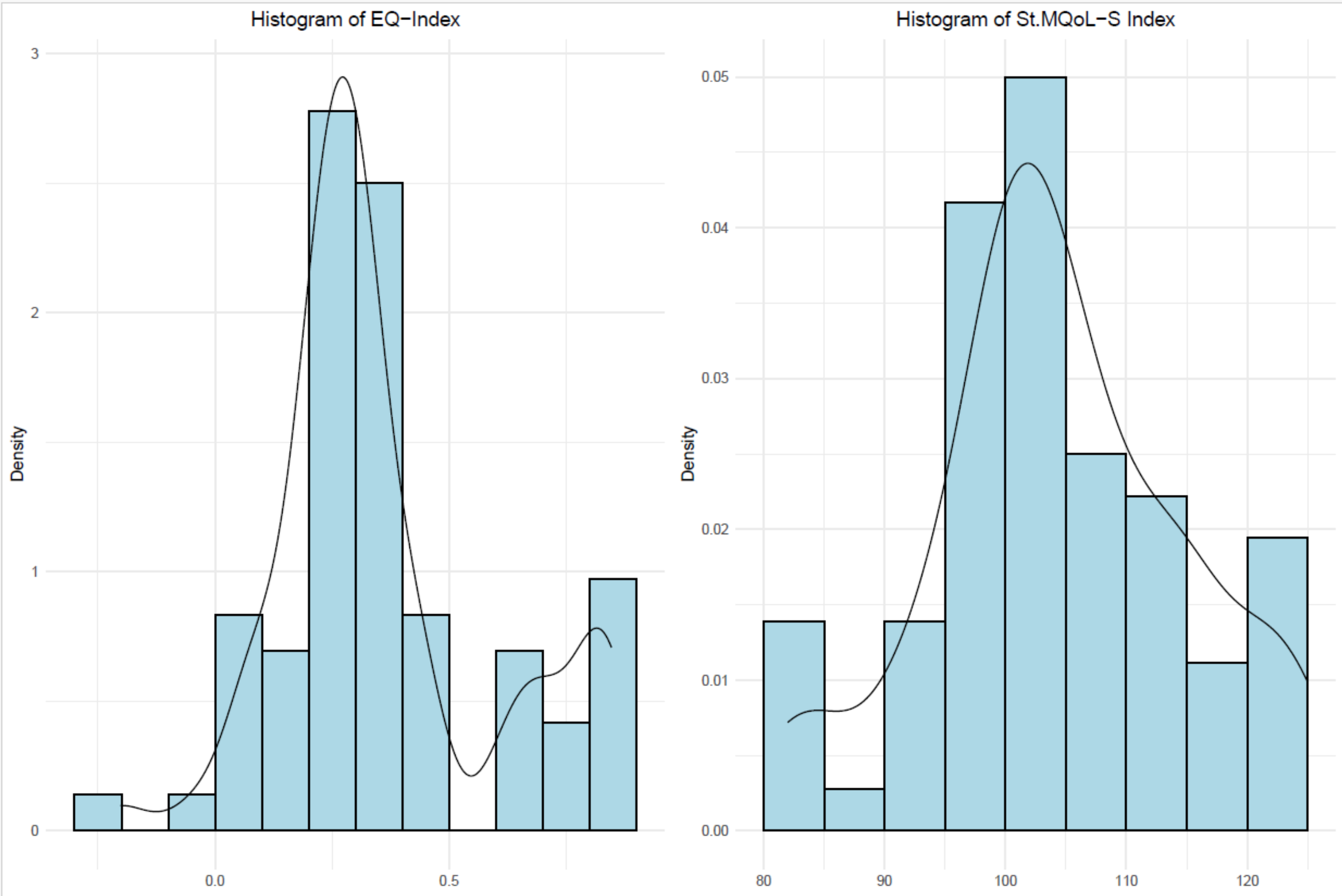

Figure S2: Scatterplot matrix between EQ-5D utility scores and domains of St. MQoL-S total scores

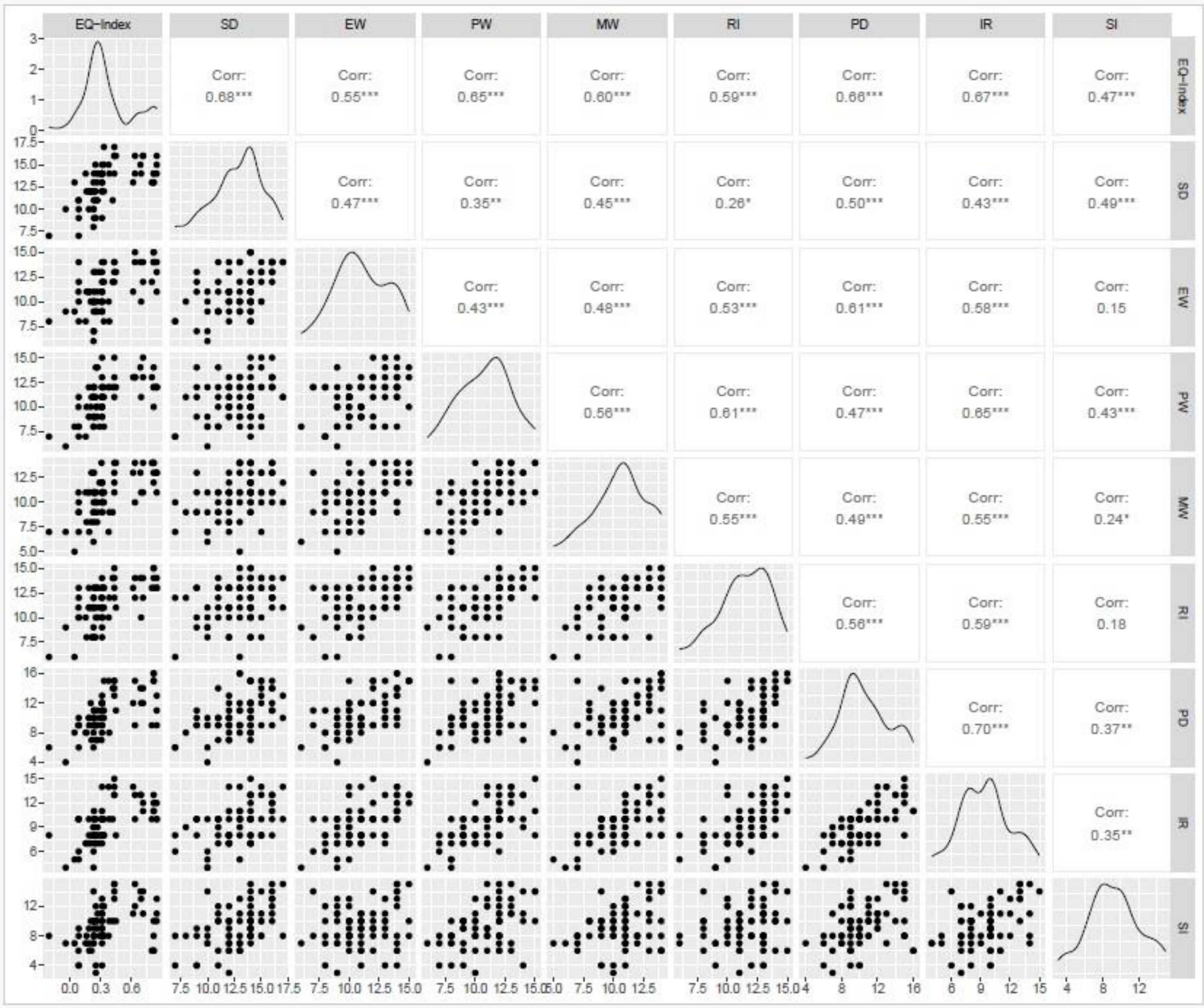

Figure S3: Scatterplot between observed and predicted EQ-5D utility scores (Model 1)

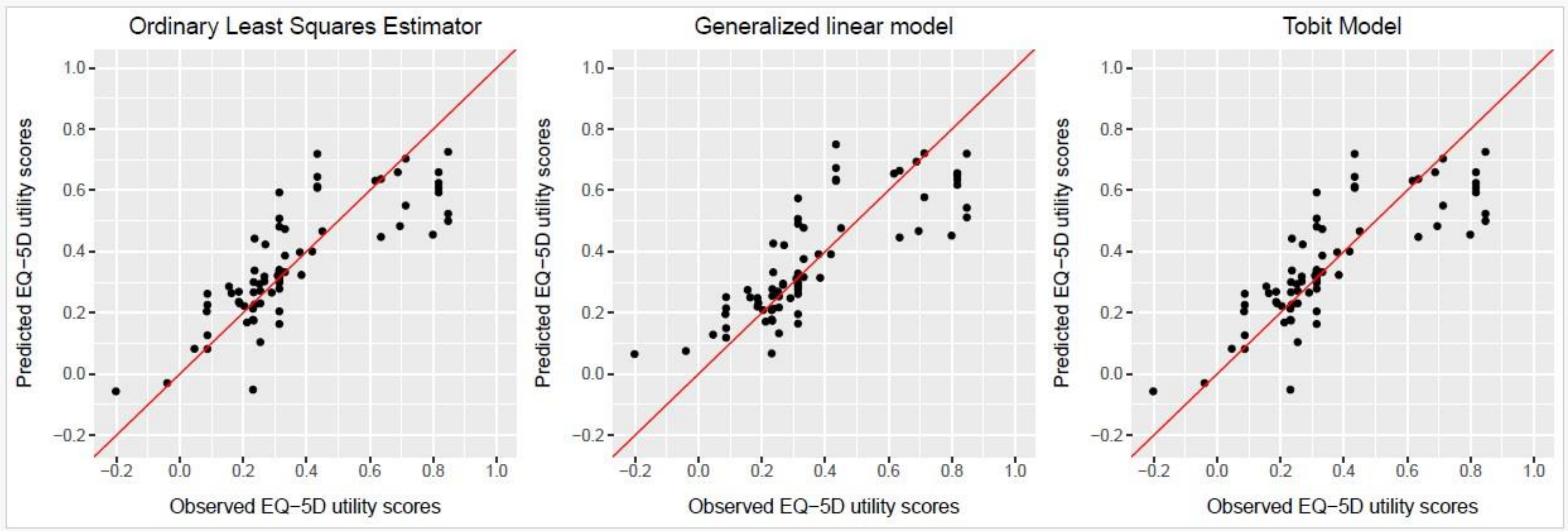

Figure S4: Scatterplot between observed and predicted EQ-5D utility scores (Model 2)