Psychometric Validation of the Traditional Chinese Version of the Early Onset Scoliosis-24 Item Questionnaire (EOSQ-24)

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Introduction

Early Onset Scoliosis (EOS) can be defined as an abnormal lateral curvature of the spine that presents before 10 years of age.[1] Possible causes include congenital vertebral anomalies, neuromuscular, syndromic or idiopathic. Due to the large growth potential, the earlier the onset of scoliosis, the worse the final curvature will be.[2] Hence, if left untreated, the natural course of EOS include severe spinal deformity, compromised respiratory function and pulmonary failure[3,4], cardiac disease, and increased mortality.[2,5] EOS should thus be managed early once diagnosis has been made. Unsatisfactory treatment can undoubtedly impose significant mental, physical and financial burdens on patients as well as their parents or caretakers.[6]

In order to properly assess the impact of the disease process and treatment provided, it is desirable to not only monitor by clinical and radiographic parameters, but to consider the importance of quality of life as a measure of treatment outcome. A disease specific instrument for assessment of the EOS patients will be useful to assess patients' functional and emotional status, and equally important the effect on parents and caretakers. The Early-Onset Scoliosis Questionnaire (EOSQ) was developed [7] to reflect the important issues of EOS patients and their caregivers, and it was found to be capable of measuring the quality of life of EOS children[6] with construct validity showing unique domains measuring intended issues of interest. The more commonly used version, Early-onset Scoliosis 24-Item Questionnaire

(EOSQ-24), was shown to be able to demonstrate differences in quality of life of EOS patients pre- and post-operatively, and it appears to be an appropriate outcome measure for comparing treatment options.[8]

Different ethnicities have variable presentations with regards to spine pathologies[9] and may also have an effect on symptom severity, treatment indications and outcomes. Management decisions should thus be tailored towards the respective population group. Given the size of China's population, there are hundreds and thousands of EOS patients who do not yet have any objective assessment scale to measure treatment outcomes. It is necessary to have a standardized method of assessment of patient or care-taker-perceived outcomes to compare treatment modalities and determine the best management option for patients with regards to health-related quality of life. This helps clinicians decide on interventions that are evidence-based rather than based upon clinician belief. This provides EOS patients with the best quality of care. Hence, the aim of this study is to translate the EOSQ-24 questionnaire from its original English version to traditional Chinese, as well as to maintain the characteristics of the original property of EOSQ to facilitate quality of life assessments of Chinese EOS patients.

Materials and Methods

Subjects and Setting

A convenience sampling of Southern Chinese patients diagnosed with EOS were recruited at the Duchess of Kent Children's Hospital in Hong Kong during the months of November 2015 to January 2016. Those who were excluded from recruitment included patients diagnosed with scoliosis at the age of 10 or over, non-Chinese, and those parents or caregivers who were illiterate and could not understand traditional Chinese. Ethics approval was obtained from the institutional review board of the University of Hong Kong/Hospital Authority Hong Kong West Cluster (reference number: UW 15-596).

Subjects' parents/caretakers were approached by research personnel, and those who consented answered a structured questionnaire which consisted of the translated, traditional Chinese (Hong Kong) version of EOSQ-24 questionnaire (**Appendix A**), followed by a Likert scale for assessing the ability of understanding the just completed EOSQ-24 (traditional Chinese Hong Kong) by the parents/caregivers. The traditional Chinese version of the Child Health Questionnaire – Parent Form 50 (CHQ-PF50) was then administered for validation.

Demographic and clinical data at the time of visit were collected. The type of EOS diagnosed was indicated as congenital, neuromuscular, neurofibromatosis, syndromic, spinal cord anomaly and idiopathic. The age at diagnosis of EOS, the age at recruitment and gender of patients were recorded. Without prior knowledge of the conduction of questionnaires, a spine surgeon performed the consultation and radiographic measurement as usual. Cobb

angle measurements were performed on whole spine posteroanterior radiographs taken in either standing or erect in sitting posture. The curve magnitude (classified as either ≤30° or >30°) and location of the curvatures were recorded. This curve magnitude was chosen because it was likely the threshold where clinicians intervened with brace treatment. Other clinical parameters also included the type of treatment modalities: under observation only, having bracing, completed bracing, post-operative, with planned surgery, and undergoing growing rod distractions. Whether the patients could walk without aids, required walking aids like crutches, or was non-ambulatory were noted.

Translation and Cross-culture Adaptation

The original version of EOSQ-24 is in American English. It was translated into traditional Chinese (Hong Kong) following one of the internationally accepted translation techniques[10], which consisted of a double forward translation and a single back translation. The translations were performed by independent professional non-physician translators to make as suitable to lay-persons as possible. After the first forward translation, the translated traditional Chinese version of EOSQ-24 was reviewed by a panel of health-related professionals (consisting of at least one spine specialist) who were from the local Hong Kong population. It was then back translated into English by a professional non-physician translator who had no access to the original questionnaire. The final forward translation was carried out

by an independent professional non-physician translator, and the final version of the translated and culturally-adapted EOSQ-24 in traditional Chinese (Hong Kong) was finalized and approved by the review panel.

Study Instruments

EOSQ-24

The EOSQ-24 is a disease- specific questionnaire with 24 questions with 11 separate sub-domains and 3 domains. The sub-domains were general health, pain, pulmonary function, mobility, physical function, daily living, fatigue, emotion, parental burden, financial burden, and satisfaction. The domains were patients' quality of life, burden, and satisfaction. There were five responses from point 1 to 5 per item in a ranking style with relevance to the questions. All 11 sub-domains scores were calculated as (average points of items for sub-domains – 1)/4*100, rescaling the score metric from 1-5 to 0-100. All 5 domain scores were calculated in the same matter. The EOSQ-24 total score was the average of 11 sub-domain scores. There was also a five-point Likert scale for parents/caregivers to rate regarding the clarity and understanding the translated EOSQ-24.

CHQ-PF50

The CHQ-PF50 was designed to assess the functional status and psychosocial

well-being of children 5 years and older and relative burden of care on parents.[11] The CHQ-PF50 included 13 domains and 2 summary scores with 50 items. The domains were Global Health, Physical Functioning, Role/Social Limitations-Emotional/Behavioural, Role/Social Limitations-Physical, Bodily Pain/Discomfort, Behaviour, Mental Health, Self-Esteem, General Health Perceptions, Parent Impact-Emotion, Parent Impact-Time, Family Activities, and Family Cohesion. According to CHQ-50PF scoring manual[12], 10 selected domains score were aggregated into two summary scores: Physical Summary Score and Psychosocial Summary Score. In brief, each selected domain score was standardized using a z-score transformation which the difference between domain score and general population mean was divided by standard deviation of general population. Computation of Physical Summary Score was obtained by multiplying each selected CHQ-PF50 scale z-score by its respective physical factor score coefficient and summing the ten products. All domains and summary scores ranged from 0 to 100.

Statistical Analysis

Descriptive statistics including mean, standard deviation (SD) and percentage of floor and ceiling of domain and total scores were calculated. At least 15% of patients achieving the lowest or highest possible score was considered as present for floor or ceiling effect, respectively.[13] By using Pearson's correlation test, the construct validity of the EOSQ-24

domain was assessed against the CHQ-PF50 domain scores holding similar constructs.

The internal consistency was assessed by Cronbach's alpha using a value >0.7 to indicate adequate internal consistency and reliability.[13,14] The sensitivity of the EOSQ-24 score was determined by performing known group comparisons by effect size, independent t-test and analysis of variance, where appropriate. Cohen's effect size was calculated as the difference between mean scores, divided by pooled SD. Comparisons of known clinical groups were: (i) Observation treatment versus bracing or surgery; (ii) Observation treatment versus bracing only; (iii) Bracing versus surgery; (iv) Curve magnitude: Cobb angle ≤30° versus >30°; (v) Ambulatory versus non-ambulatory; and (vi) Various types of EOS- Congenital versus neuromuscular versus syndromic vs idiopathic. As EOS of neurofibromatosis (2%) and spinal cord anomaly (3%) nature were of such small proportion of the study population, they were excluded in the analysis of clinical diagnosis group comparisons.

Data analyses were conducted using SPSS Windows 23.0 (IBM SPSS Inc., Chicago, IL, USA). P-value<0.05 was considered statistically significant.

Results

A total of 100 patients (60 females, 40 males) who were diagnosed with EOS were recruited, and their parents/ primary caregivers were asked to fill in the translated EOSQ-24

and CHQ-PF50. The response rate was 100% and all questionnaires were filled in the clinic without any missing responses. This studied population was heterogeneous, consisting of various aetiologies for EOS, spinal curvature of different magnitudes (Cobb angles), undergoing different stages of treatment and ambulatory status (**Table 1**).

Understanding of translated EOSQ-24

A 5-point Likert scale was used to assess the understanding of the Chinese EOSQ-24 from the parents/caregivers who filled the translated questionnaire, giving an indication of its clarity. There were 66% and 10% of the parents/caregivers who answered 'Agree' and 'Strongly Agree' respectively on their ability to understand the EOSQ-24 questionnaire. Only 15% answered 'Neither Agree/Disagree', and 9% for 'Disagree'. No subjects chose 'Strongly Disagree'.

Internal Consistency

Table 2 presented the mean, standard deviations, floor and ceiling effects of the EOSQ-24 item, sub-domain, domain and total scores. The Cronbach's coefficient was calculated for the analysis of reliability (**Table 2**). The Cronbach's coefficient of the total EOSQ-24 score was 0.896, indicating a very good reliability. The internal consistency of the three domains of EOSQ-24, namely Patient Quality of Life, Family burden and Satisfaction,

was excellent (Cronbach's α: 0.829-0.919). The range across all sub-domains was 0.589 to 0.930.

Ceiling and Floor effects

For the study population, the mean sub-domain scores of EOSQ-24 ranged from 57.9 (General Health) to 84.4 (Pulmonary Function), and the mean scores of individual items ranged from 2.9 (Q1 of General Health) to 4.4 (both Q5 & 6 of Pulmonary Function). A total of 10 items were not skewed, since Item 1, 12, 13, 14, 17, 18, 19, 23 and 24 had a median score of 3 and the median for Item 10 was 3.5 (50^{th} and 51^{st} response values were 3 and 4 respectively). The rest of the items were left-skewed, with Item 5, 6, 7, 8 and 9 being highly left-skewed, indicating responses pointing towards normal mobility, physical and pulmonary function of this study population. With the absence of any right-skewed items, a floor effect was found in 0% to 26% of patients and a ceiling effect in 0% to 71% of patients (**Table 2**).

Validity

As summarized in **Table 3**, the total score of EOSQ-24 had significant correlations ($p<0.001$) with all domains of CHQ-PF50. Also, the three individual EOSQ-24 domains correlated significantly with all CHQ-PF50 domains. The sub-domain scores of EOSQ-24 correlated significantly ($p<0.001$) with those of CHQ-PF50, particularly strong correlation in

the sub-domains of Pain, Physical Function and medium strength of correlation in General Health, Mobility, Parental Burden and Emotion. The Satisfaction domain of EOSQ-24 had strong significant correlation with the domain of Self-esteem of CHQ-PF50. A good construct validity was demonstrated and depicted in **Table 3**.

The discriminative validity could be indicated by the sensitivity of the EOSQ-24 in differentiating known clinical groups as displayed in **Table 4**. The EOSQ-24 was able to detect statistical differences in its total score between patients who were ambulatory versus non-ambulatory, as well as between EOS patients of various etiologies. There was statistical significant differences in the domain Family Burden and sub-domain Emotion and Financial Burden for patients undergoing bracing as compared to those who had surgery, despite the total score difference being not statistically significant (p=0.092). Other clinical groups could not be differentiated by EOSQ-24 based on the statistical value, however, there were sub-domain scores which could be suggestive of being important when comparing various stages of treatment and the magnitude of the spinal curvature.

Discussion

This is the first study to translate and culturally adapt the EOSQ-24 for the Chinese population. This is a necessary first step to provide a standardized outcome questionnaire for Chinese EOS patients to allow for outcome comparisons and for determining the best

treatment options based on health-related quality of life assessment. Our results show that this translated questionnaire is well accepted by the study subjects and have demonstrated good construct validity and excellent reliability for comparing different diagnoses, ambulatory status and treatment modalities. We were also able to determine significant differences between domains of Emotion, Family Burden and Satisfaction among different EOS diagnoses.

Patients who are diagnosed with EOS require early treatment. Outcome assessment does not only include monitoring severity of spinal curvature, but also quality of life assessments of patients and their primary caretakers. It is demonstrated that scoliosis and resultant thoracic insufficiency may have a profound impact on patient's quality of life.[15] In fact, unlike adolescent idiopathic scoliosis, at a young age, EOS patients face the risk of poor thoracic development leading to associated pulmonary morbidity and early mortality.[3,4] Patients with EOS may have an equally poor quality of life as children with severe asthma or cardiac disease.[6,16]

Previous analysis of the generic instrument CHQ alone in patients with thoracic insufficiency syndrome (TIS) is unable to reveal a moderate or large degree of change of quality of life before and after vertical expandable prosthetic titanium rib (VEPTR) surgery.[16] Despite the lower physical domain score, the scores in psychosocial domains in patients with TIS are shown to be similar to those in healthy children.[16] This raises the question of whether a

generic instrument like CHQ has the adequate responsiveness and sensitivity to detect specific changes in quality of life found in this particular population of patients.

For EOS patients, EOSQ appears to be a more appropriate and reliable instrument, as it is a patient-based quality of life measure. Every step of its development involved the primary caregivers of patients, as well as input from health professionals.[6] Item and domain creation were driven by patient-based issues of concern that reflect quality of life and caregiver burden, through various stages of treatment and clinical severity.[6] The EOSQ-24 is shown to demonstrate differences in quality of life before and 6 months after surgery (VEPTR, traditional growing rods and Shilla) and can be an appropriate measure of outcome in comparing treatment options for EOS patients.[17] Hence, for the Chinese population, it is necessary to translate the EOSQ-24 into Chinese and validate it using the CHQ-PF50, in order to project the quality of life of these young patients from their parents/caregivers' point of view.

This study has found good internal consistency and thus reliability for the total score and domain scores for the translated EOSQ-24. The internal consistency of the Chinese EOSQ-24 (Cronbach's α: 0.896) is found to be comparable with other translated EOSQ-24, namely the Turkish (Cronbach's α: 0.909) and the Spanish (Cronbach's α: 0.897) versions.[18,19] The Chinese EOSQ-24 is also found to good construct validity, with strong correlation of scores with all domains of CHQ-PF50. Moreover, the translated EOSQ-24 demonstrates sensitivity

in detecting differences between subjects who have different diagnosis and ambulatory status. It is interesting to recognize that the differences are significant in the domain Satisfaction ($p<0.001$) as well as Family Burden ($p=0.014$) among various diagnosis groups, together with multiple sub-domains. Among patients undergoing different treatment, EOSQ-24 is sensitive in differentiating between patients undergoing bracing as compared to those who undergo surgery by the domain of Family Burden, accounted by the Emotion and Financial Burden sub-domain. Even though the EOSQ-24 does not capture a statistical significant difference between patients under observation versus other treatment modalities, nor differentiating patients with different severity of curvature magnitude, the instrument still detects the relevant sub-domains which play a strong role suggestive of differentiating among the groups. For instance, for patients with different magnitude of curvature (Cobb angle $\leq 30°$ versus $>30°$), sub-domain Pulmonary Function has a p-value of 0.067 (effect size=0.367), whereas patients with bracing versus those with surgery done, sub-domain Pain has a p-value of 0.082 (effect size=0.408).

One limitation of the study is that it was performed in a single tertiary center in Hong Kong. Its generalizability to the entire Chinese population is unknown. It is fortunate that there is only one common written language that is shared by all Chinese populations, with the traditionally written characters used in Hong Kong, Macau and Taiwan, and simplified characters used in Mainland China. The traditional Chinese version can be converted to

simplified Chinese characters for application to the rest of Mainland China, but some minor modification may be necessary to suit the local linguistic preferences on certain terminologies. The modification is expected to be minimal since there are no cultural differences between these populations. Nevertheless, both character versions are usually understandable for those who read Chinese.

This is thus the first study to translate and culturally adapt the EOSQ-24 for the Chinese population by native health-related professionals. The translated questionnaire is well accepted by the parents/caregivers and demonstrates good construct validity, excellent reliability, and also sensitive for discriminating subjects with different diagnosis, ambulatory status and treatment modalities. This EOSQ-24 questionnaire is successful in capturing significant differences in the aspects of Emotion, Family Burden and Satisfaction among different diagnoses under EOS. These are domains which cannot be effectively measured by clinical parameters alone and are often difficult to gauge by clinicians. Yet, they are important contributing factors towards the quality of life of EOS patients and burden of caregivers. In addition, it can be an important disease-specific instrument for measuring outcomes of treatment specific for Chinese EOS patients. It will be ideal as the next step to test the responsiveness of the Chinese EOSQ, and with a much larger sample size to reveal more effective domains successfully.