



CHAPTER V

Discussion and Conclusion

The goal of this study was to develop and undertake initial validation analyses of the QoLMEAL as a tool for measuring the SLE-specific pediatric HRQOL with parallel child and parent reports. The results of this study provide preliminary evidence of the reliability and validity of the QoLMEAL in this population. Our instrument was designed for teenager with SLE aged 10-18 years. We assembled the questionnaire from an extensive review of the existing childhood SLE and HRQOL literature and HRQOL measures, and the information from the health-care providers who experience in managing SLE adolescents. Subsequently, we showed that the instrument is valid for childhood SLE because it possesses construct validity, content validity, and internal consistency. Though health-care professionals chose the items at initial, the involvement in an item generation as well as revision of the patients and parents followed through the study. Crucially, they were asked to give comments on the items or suggest additional items if they felt important areas were omitted.

Values of ceiling or floor effect above 20% are considered high.(69) Almost a half of our patients selected a minimum score of zero in finance domain (self report), so the differences might not be detected between the groups with different severity by these three questions in finance domain. While there was the floor effect in daily activity, family, and social domain, this did not invalidate the questionnaire as the items contribute to the content validity. However, most of our patients were recruited from the out-patient clinic and were not in hospital wards, so they had either milder disease or controlled disease confirmed by the result of ECLAM scoring assessment. Therefore, more studies on other patient populations are needed.

In this preliminary study, the questionnaires with incomplete data were removed from the analysis of involved domain as well as total scale. 16.4% of self reports and 23.6% of parent reports were incomplete. The most frequently missing data was the item

in social domain both adolescent- and parent-report [S10] and in treatment domain parent-report [T3].

[T3], โรคของเด็กเป็นโรคที่รักษายาก (My child's illness is hard to treat.)

[S10], เป็นเรื่องยากที่ฉัน / เด็กจะช่วยเหลืองานของ พ่อ แม่หรือผู้เลี้ยงดู (It is hard for me / my child in help with housekeeping.)

We realize that the complete case analysis in the study reduces the sample size and may produce misleading results. No reason of leaving the questions unanswered was reported. We speculated that the parents might be uncertain how their children's SLE behaved or whether SLE was under good control with their current treatment. The children might barely be asked for any help from their parents probably due to some concern about potential SLE flare.

With regard to the item-scaling analysis, the items comprising each domain mostly correlated with each other (convergent validity) and not correlated too highly with other domains (discriminant validity). Convergent validity is supported if an item correlates moderately ($r \geq 0.3$) with the domain it is hypothesized to belong to. However, some items in daily activity domain adolescent-report [DA1, DA4, DA5, DA6, DA8] and parent-report [DA5] showed only small correlations not only with other domains but also with its own domain.

[DA1], การเดินบนพื้นราบภายในบ้านเป็นเรื่องลำบากสำหรับฉัน

(It is hard for me to walk.)

[DA4], การติดกระดุมเสื้อเป็นเรื่องลำบากสำหรับฉัน

(It is hard for me to button up my shirt.)

[DA5], การให้ช้อนตักอาหารเข้าปากเป็นเรื่องลำบากสำหรับฉัน / เด็ก

(I have trouble eating with a spoon)

[DA6], การหวีผมเป็นเรื่องลำบากสำหรับฉัน

(It is hard for me to comb my hair.)

[DA8], การอาบน้ำเป็นเรื่องลำบากสำหรับฉัน

(It is hard for me to take a bath or shower.)

The poor correlations suggest they might be measuring the different things from their domain. Interestingly, all of the above items connote the very basic daily activities without any physical exertion. Although SLE is associated with pain, bone aches, weakness, and stiffness in these children(45), our patients whose disease mostly was mild or under control might barely experience difficulty in doing these activities. These items might be omitted from the questionnaire if the results of our final report show the same poor correlations.

In social domain adolescent-report, one item [S4] had an equal correlation with its own domain and the emotional health domain.

[S4], ฉันอายจนไม่อยากจะไปนอกบ้าน

(My embarrassment makes me avoid social situations.)

The [S4] item probably reflects both in the social and emotional concept. The factor analysis also implied that [S4] was as moderately associated with social as with emotional health, and that either factor could influence the value of [S4].

To test the internal consistency, Cronbach' coefficient is used for our study. It is a measure based on the correlations between different items on the same scale (or the same domain). Coefficients above 0.7 are generally regarded as acceptable for psychometric scales.(70) The high value of Cronbach's α (0.94) for the entire scale indicates that the QoLMEAL has good internal consistency. With the items in each domain, Cronbach's α ranges from 0.68 to 0.87 which is comparable to the values for other validated instruments such as SMILEY (α for domain, 0.7 – 0.8).(38)

The instrument emphasizes the adolescent's subjective perceptions in the context of SLE. Although it is ideal for children and adolescents to evaluate their own QOL, there are limitations such as unreliable or invalid results based on their varying degrees of long-term perspective, memory, age, and communication skills.(60) Parents' reports are extremely useful in these cases and their perception affects children's health care utilization.(71) Parental expectations, feelings of guilt, health, mental status, QOL, and values may influence their reports and lead them to perceive their child's QOL as being poor.(60) The level of parent-child agreement varies and is dependent on the

measurement method, domains studied, and children's/parents' physical and mental health status.(72-74) The observed moderate correlation between the adolescent and parent reports in our study further supports the need for parallel child and parent reports to obtain both perspectives.(53, 71) There was no relationship between some items (shown as below) in daily activity domains adolescent- and parent-report.

- [DA1], การเดินบนพื้นราบภายในบ้านเป็นเรื่องลำบากสำหรับฉัน / เด็ก
- [DA4], การติดกระดุมเสื้อเป็นเรื่องลำบากสำหรับฉัน / เด็ก
- [DA5], การใช้ช้อนตักอาหารเข้าปากเป็นเรื่องลำบากสำหรับฉัน / เด็ก
- [DA6], การหิ้วนมเป็นเรื่องลำบากสำหรับฉัน / เด็ก
- [DA8], การอาบน้ำเป็นเรื่องลำบากสำหรับฉัน / เด็ก

Only small correlations between the daily activity domains of the adolescent and parent reports suggest either the different perspectives / perceptions of the child and the parents in the aspect of daily activity or the domain measuring different things.

That our instrument only correlated weakly with the SLE activity indices is consistent with the previous reports in adult and children. Our patients with more active disease reported a poorer quality of life across the physical health, daily activity, treatment domains, whereas all domains were not able to discriminate between patients with or without damage. However, most damage found in our patients was mild cataract which was asymptomatic and did not interfere in their vision. PedsQL Generic module did not correlate with SLEDAI.(42) Only mild correlations were found between the limitation domain scores of the child SMILEY report and the SLEDAI, PGA, or SDI.(38) There was poor correlation of the SLEQOL and the SF-20 with SLEDAI(18, 27) and the SF-36 with BILAG.(30) On the other hand, other investigators have found that Child Health Questionnaire correlated with SLEDAI and SDI(40) and the SF-36 correlated with SLEDAI(27), the global disease activity measured by BILAG(20) and SLAM(16). This suggests that lupus activity and damage do not consistently perturb the QOL. One possible explanation is that local factors and personality have more a direct effect on the subjective perception of QOL than life circumstances or disease.(75)

Our instrument demonstrated some discriminative properties, as scores increased going from low to high disease activity. Increased disease activity often causes limitation of activities and fatigue as well as requires more frequent doctor visits and more medications to control the disease. Therefore, it is not surprising to see a greater discriminatory trend in the physical health and daily activity domain as well as treatment domain. The study population comprised mainly high-school attending teenage girls, with low disease activity and disability followed at tertiary care centers, which may limit generalizability. An increase in sample size might likely results in significant discrimination in other domains.

We have documented the steps we have undertaken to construct a SLE-specific QOL instrument, and we showed that it possesses construct validity, content validity, and internal consistency. Thus, the validity of our instrument has been established. Full validation is awaited; work that remains to be done include resolving its role in clinical trials and routine practice and confirming its applicability in different populations. Item reduction in the questionnaire may be undertaken using information gained from the principal component analysis, patient feedback, and clinical decision when our sample size is large enough. Test-retest reliability should be assessed in the near future. Our instrument may ultimately prove itself to be a useful adjunct to clinical practice and research, providing valuable insight to the impact of SLE on the overall QOL of the adolescents.

Conclusion

QoLMEAL is a valid instrument for measuring the quality of life of Thai adolescents with SLE. However, several limitations should be considered when interpreting our findings. Subjects were drawn from a convenience sample. The study population was fairly homogeneous, comprised mostly of out-patient subjects. Qualitative studies were not performed due to the time limitation of the patients and parents attending the out-patient clinics and a lack of the experiences in focus group discussion or deep interviews of the investigators. More than 20% of our questionnaires were incomplete in this preliminary study but the complete case analysis remained to be

used. Concurrent validity was not evaluated due to unavailable validated quality of life measure for Thai children or adolescents. The criteria used to categorize the subjects for the known-group validity evaluation may not have been sufficient to allow for the scales to detect differences between groups. Grouping the subjects by a different criterion, such as a relevant clinical measure, may approve the findings for its known-group validity