CHAPTER V
DISCUSSION

1. Research Sample

The sample data were collected from patients with cleft lip and/or palate records in the Orthodontic Department, Faculty of Dentistry, Khon Kaen University according to the inclusion and exclusion criteria.

The present study comprised of 105 subjects with slightly greater numbers of females (59, 56.2%) than males (46, 43.8%). Most of these subjects were in the age group between 5 and 11 years (41.9%) in both sexes and presented with the unilateral cleft lip and palate defect (71.4%).

Even though a large number of subjects was collected, only 105 subjects were met the selection criteria. This was mainly due to the lack of cleft samples who had received their final treatment plan. Currently, with a better access for receiving of information and appropriate treatment, most of the new cleft patients are the newborns or still-growing patients. These patients undoubtedly require a long period of time, until their growth is completed or almost completed, before the final treatment plan can be made. This factor may limit the number of the subjects in this study.

2. Research Sample Compared with the Previous Study[22]

Compared with the study of Singhawannakul[22], characteristics of the samples in this study were similar in which majority of those subjects were females (54.6%), aged between 8 - 11 years (52.1%), and presented with unilateral cleft lip and palate defect (63.0%).

The percentage of subjects who had participated in Singhawannakul's study was 43.8%. Most of these previously participating subjects were female (60.9%) and half of them also had an age less than 12 years (50.0%). Although the lowest age limit that met the inclusion criterion of having a later final orthodontic treatment plan implemented or completed in this study was 5 years old which was less than 8 years

of the lowest age limit in the previous study, there were only 4 subjects who had an age less than 8 years. This could be considered as only a small amount. Moreover, since the FOSP was intended to be applied to all age ranges, these samples were also included.

3. Actual Orthodontic Treatment Plan of the Samples

The proportion of subjects who required orthodontic treatment only was larger than that of subjects who required additional orthognathic surgery (55:45). This might indicate the nature of the acceptance for additional orthognathic surgery at the Cleft Palate Center as well. The percentage of subjects who were predicted to receive orthodontic treatment combined with orthognathic surgery was approximately 45%, conforming with 46.2% as estimated by Singhawannakul.[22] Approximately 20% of male subjects with UCLP received orthodontic treatment combined with orthognathic surgery which was comparable to 25% as reported by several other studies.[13, 33, 89]

Tendency for requiring additional orthognathic surgery was evident in cleft deformities that involved both primary and secondary palates (CLP), including both UCLP and BCLP, while only orthodontic treatment seemed to be sufficient in the isolated CL and CP groups. This finding might relate to the severity of cleft involvement.[69, 87] The more severe the cleft involvement, the more additional orthognathic surgery was likely required. Previous studies also showed that additional orthognathic surgery was frequently necessary in subjects with BCLP due to the difficulty in obtaining good occlusal relationship and desirable esthetics via orthodontic treatment alone.[88, 104]

According to Kiattavarncharoen and Ouychai[74], the Skeletal Class III problem in Thai noncleft females commonly originated from both retrognathic maxilla and prognathic mandible while only mandibular prognathism was involved in the male nonclefts. This finding implied that severity of Skeletal Class III problem might increase in both sexes due to maxillary retrognathism that is usually observed in the repaired clefts. Interestingly, in the BCLP group of the present study, it was obvious that additional orthognathic surgery was mostly anticipated in males rather than females. Longer period of the mandibular growth in males might play a role. It was also possible that adaptive retrusion of the mandible as reported by previous studies[75-77] might not have occurred in males with

BCLP in this study. Instead, it might occur in males with UCLP. Longitudinal study with larger samples would clarify this postulation.

4. Reliability of the Cephalometric Measurements

All three cephalometric measurements required for the FOSP which are ANB (degree), U1-APog (mm), and L lip-Nperp. (mm) were collected twice by one examiner. Excellent reproducibility was observed in all three parameters (ICC values were more than 0.80).

5. The Formula for Orthodontics and Surgery Prediction (FOSP)

Rigid criteria for judgment of the orthognathic surgery need have not been established. Therefore, Singhawannakul developed the FOSP with an aim to facilitate treatment planning in the cleft patients.[22] This formula could predict whether additional orthognathic surgery should be incorporated in the treatment plan. Both reliability and validity in the use of the FOSP as a clinical tool aiding in treatment planning, were evaluated in this study.

5.1 The critical D score

To obtain the optimal cutoff D score that could delineate between the two types of treatment need, orthodontic treatment only or additional orthognathic surgery, the receiver operating characteristic (ROC) curve was used to identify the critical D score. Since the FOSP was originally intended to be used as a diagnostic tool for assessment or prediction of orthognathic surgery need, a high specificity to rule in this condition was necessary while sensitivity must be optimized as well. The critical D score of the FOSP in this study was 0.64775, providing the most desirable percentages of both sensitivity and specificity. A higher D score beyond this point indicated the prediction of the need for orthognathic surgery while the lower score signified orthodontic treatment alone.

5.2 Reliability and validity test of the FOSP

The critical D score at 0.64775, reliability of the FOSP was assessed by considering agreement between actual treatment plan and the predicted treatment plan interpreted from the FOSP score. According to standards for classification of the strength of agreement as proposed by Landis and Koch[101], kappa value of 0.50

indicated only a moderate strength of agreement after agreement by chance was taken into account.

To evaluate the validity of the FOSP, the most accurate and proper treatment plan must be used as a standard or reference. An ideal treatment plan should provide good post-treatment stability and satisfy patient's appearance after completion of the treatment. Nevertheless, until now, there are still no rigid criteria that can clarify whether orthognathic surgery should be included in the treatment plan. This is because the need for the surgery is a perceived need with various decision factors having influence, as mentioned earlier.[33, 92] Validity of the FOSP in this study was assessed using the actual treatment of each subject as a reference. No post-treatment records were analyzed to evaluate the treatment success or propriety of the original treatment plan. Although long-term stability of, as well as patient's satisfaction with dento-skeletal outcomes should be evaluated when ideal treatment planning is desired, the actual treatment plans were established or supervised by each individual's orthodontist who had experienced in treating cleft deformities for more than 10 years at the Department of Orthodontics, Khon Kaen University. The actual treatment plan in this study was, therefore, a combination result of judgment from both patient and experts.

Sensitivity and specificity of the FOSP for prediction of orthognathic surgery need were 68% (68.6 - 90.1% of 95% CI) and 81% (52.9 - 80.9% of 95% CI), respectively. High percentage of specificity indicated that the FOSP tended to have an ability to identify candidates for additional orthognathic surgery. This could subsequently reduce the amount of subjects who would be misclassified as needing the surgery. Nevertheless, wide ranges of confidence intervals in both sensitivity and specificity were of concern as they might reflect an inadequacy of both surgery and non-surgery subject numbers. Larger samples are required to ensure these findings.

The percentage of correctly classified subjects (accuracy) was 75.2% which was less than 83.2 % estimated in the initial development of the FOSP.[22] It was possible that the percentage of accuracy in this study was determined using the actually received treatment not the recommended treatment from an expert as in Sinhawannakul's study. The predictive merit of the FOSP in Singhawannakul's study was also questionable since it was based on the predictive capabilities of only one

expert clinician, and the findings of the study diluted by the large mix of subjects by age and cleft condition. Compared with a study of Nollet et al.[21], the prediction models for cephalometric outcome at age 18 years was developed from the cephalometric values at age 9 years to assess the need for surgery in 40 UCLP subjects, using logistic regression analysis. Sixteen hard and soft tissue cephalometric variables (six skeletal, two dental, and eight soft tissue profile variables) were used in their study from which four variables were determined as the most relevant cephalometric values for surgical prediction. These were SNA, SNPog, soft tissue ANB and soft tissue ANPog. Comparing the predicted treatment need with the treatment recommended from three experts, the need for additional orthognathic surgery at adulthood was correctly predicted with 85% accuracy. However, determination of the predictive value from the same population used to construct the predictive model, as performed in the studies of Nollet et al. and Singhawannakul, should be taken into account as it might possibly increase a positive bias of predictive estimation. Additionally, the number of subjects in Nollet et al.'s study, especially in the surgical group (11 subjects), was limited and differences in study population traits must be considered.

The accuracy of the present study was also less than the 92.0% predictive accuracy of the treatment prediction equation developed by Stellzig-Eisenhauer and colleagues.[97] However, their study was performed for adult Class III nonclefts subjects, not expecting any possible favorable jaw growth, while 71.4% of subjects in this study were aged less than 15 years. Moreover, existence of cleft deformities might variously affect later development of the subjects, increasing the difficulty in predicting the later treatment that would be needed. The interval between timing of treatment prediction and timing of a definitive treatment plan decision which is usually given at approximately 15 - 18 years of age might also play a role. The longer the interval between the time that treatment was planned and the time that definitive treatment was given, the more likely it was that there would be a discrepancy between them. This was because the originally presented conditions might have been changed through growth of the subjects themselves or other potential factors such as maxillary growth restriction after surgical repair of cleft deformity.

Considering the D score and the treatment prediction, the mean D score of the predicted non-surgery group was -1.58 while in the predicted surgery group, the mean D score was 2.73. These findings corresponded with those of the former study of Singhawannakul[22], the mean D score in the predicted non-surgery group and surgery group being -2.11 and 2.06, respectively. The mean values of the three cephalometric measurements used in the FOSP calculations indicated lesser values of the ANB and more positive values of the L lip-Nperp. (mm) than those of that former study in both groups of treatment need, while the U1-APog (mm) was quite comparable.[22]

In the misclassified groups, the mean D scores calculated from the FOSP were closer to those of the opposite group of actual treatment need (Table 12). The same finding was also evident when considering the three cephalometric values, the ANB (degree), U1-APog (mm), and L lip-Nperp. (mm) (Table 10). This clearly showed that using only three cephalometric values of the FOSP was apparently not sufficiently sensitive to predict the type of orthodontic treatment need in all situations, most likely for borderline cases.

5.3 Influence of an age on reliability and validity of the predicted treatment plan from the FOSP

During an intensive growth period of 8 to 16 years, an obvious reduction of the ANB angle due to reduction of the SNA angle was reported in the UCLP.[19] Other longitudinal studies also showed a decrease of maxillary prominence during aging as well.[8-10] However, individual variation of changes due to the maxillary growth inhibition must be recognized as suggested by Ross.[13] Since the D score might be affected by alteration of the ANB angle during aging, it was seen as necessary to evaluate influence of the age on reliability and validity of the predicted treatment plan in this study.

Better reliability and validity of the treatment plan from the FOSP was observed in the age group of more than 18 years ($k = 0.73$, accuracy = 88.9%). It was assumed that treatment plans decided for this older, post-adolescent age group would not need to take account of any adverse post-treatment changes due to continuing jaw growth. Nevertheless, increasing pattern of both kappa values and percentage of accuracy were not observed relative to an increasing age. Wide ranges of 95% CI in

both accuracy and kappa values were also clearly evident in all age groups. These findings seem to indicate that the age might not affect reliability and validity of the FOSP for prediction of orthognathic surgery need in this study as was proposed by Singhawannakul.[22]

5.4 Influence of gender on reliability and validity of the predicted treatment plan from the FOSP

Previous longitudinal studies of the cephalometric measurements in the UCLP subjects showed that there was no sex difference in angular and ratio measurements.[8, 21, 105] By contrast, sex difference was observed in linear cephalometric measurements.[8, 105] This referred to the U1-APog (mm) and L lip-Nperp. (mm) included in the FOSP of this study. Underdevelopment of both the maxilla and mandible had also been reported with more pronounced in females than in males.[10] Nonetheless, in this study, accuracy and reliability of the FOSP for prediction of orthognathic surgery need was quite comparable in both sexes. This finding implies that gender might not play a role on reliability and validity of the FOSP for prediction of orthognathic surgery need as well.

5.5 Influence of the cleft type on reliability and validity of the predicted treatment plan from the FOSP

According to previous studies, midfacial growth and development was likely to depend upon cleft type and severity rather than sexual differences.[77, 105] Even though the FOSP was claimed to be applicable for all types of cleft, variability of the facial growth pattern in each type and severity of cleft should be recognized. More pronounced retroclination of the upper incisors and posterior rotation of the mandible were reported in BCLP than UCLP.[63, 66] These findings suggest the need to evaluate the accuracy and reliability of the FOSP in each cleft group individually since they subsequently influenced on the U1-APog (mm) and the ANB angle, respectively.

Reliability and validity of the FOSP application according to type of cleft deformity were unable to be confirmed in this study due to limited sample size in the CL/A and CP groups. Assessment was performed in the UCLP and BCLP groups. Although the FOSP was claimed to be applicable in all types of cleft defect, it seemed that the FOSP tended to provide a better prediction of the type of treatment need in the UCLP subjects. Higher accuracy and reliability were apparent in the UCLP than the BCLP.

This might be due to the original subjects included for development of the FOSP largely came from the UCLP group (63%)[22], thus, enhancing an ability of the FOSP for prediction of the treatment need correctly in this group. Slight agreement between actual treatment plan and the predicted treatment plan interpreted from the FOSP, especially in the BCLP group, indicated that there were factors other than the three cephalometric values used in the formula that influence on the treatment decisions.

6. Clinical Application of the FOSP for Prediction of the Final Treatment Need in Skeletal Class III Oral Clefts and Noncleft Patients

According to the predetermined conditions, the FOSP would be considered as a valuable clinical tool if it provided the kappa values of more than 0.61 with the percentage of accuracy not less than 75%. The results of this study showed that although accuracy of the predicted treatment plan from the FOSP was found with satisfactory percentage (75%), only a moderate strength of agreement between the actual and the predicted treatment plan was observed in this study (k = 0.50). Even though factors that were essential in treatment planning such as maxillary midline discrepancies, facial symmetry, dentofacial esthetics, and soft tissue features had been included during the FOSP development[33, 80, 83, 97], they were not found to be significant enough to be included in the FOSP.[22] It is likely that there might be factors other than the ANB (degree), U1-APog (mm), and L lip-Nperp. (mm) that also influence on the treatment decision. These possible factors are dento-alveolar health, likely long-term stability, comparative treatment costs, and patient demands and perceptions of individual needs, with their consequent decisions about treatment. The latter factor seems to be the key confounding factors and the most difficult one to determine without communication with the patient. This is because patient's decision may be contrary to the clinician's recommendation. Moreover, the effect of an age and types of cleft condition are still ambiguous.

Based on these results, the FOSP may be useful in early prediction of the treatment plan in cleft patients, avoiding the risk from orthodontic overcompensation and prolongation of the treatment time in cases with tendency of needing additional orthognathic surgery.[19] Still, it is crucial for the clinician to incorporate all sources of patient's information, including routine clinical examination (such as existence of

functional shift, periodontal status, facial asymmetry, facial esthetics), cephalometric analysis, model analysis and the most importantly, good patient communication before making any final treatment decision. In addition, early prediction of the final treatment need provides an opportunity to identify the possible future service need in the cleft care center. This facilitates in orthodontist and maxillofacial surgeon manpower assessment and planning as well.

7. Suggestions for Future Study

Further evaluation of the effect of age and cleft type on validity and reliability of the FOSP requires a larger sample size in various age ranges and all types of cleft deformity. This may reduce the possible confounding effects of age or cleft type on the treatment need prediction.

Additionally, since the examining criteria for making the decisions between orthodontics alone and combined orthodontics and orthognathic surgery were similar to those used in the noncleft patients, it might be possible to apply this formula in the Skeletal Class III noncleft subjects as well. Further study to confirm this postulation is required.