Prediction of Final Treatment for Class III Malocclusion of Patients with Oral Clefts

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Objective: *To evaluate the value of predicted treatment plans interpreted from a previously developed Formula for Orthodontics and Surgery Prediction (FOSP) in patients with cleft lip and/or palate (CLP).*
Material and Method: *Orthodontic treatment records were collected of 105 non-syndromic CLP patients between the ages of 5 and 29 years, having Class III malocclusions of varying severity. Three cephalometric values from pre-treatment lateral cephalometric radiographs were determined for use with the FOSP. A receiver operating curve (ROC) was used to identify the optimal cut-off score for classifying predicted treatment needs, whether orthodontics alone or orthognathic surgery. The clinical merit of using the FOSP was determined by comparing predicted plans with actual treatments.*
Results: *The predictive value using a cut-off score of 0.6475 from the FOSP-for recommending the final treatment provided-was correctly classified for 79 subjects (75.2%). Sensitivity and specificity of the FOSP for prediction of the need for orthognathic surgery were 68.1% (52.9%-80.9%) and 81.0% (68.6%-90.1%), respectively.*
Conclusion: *The FOSP can be used for early treatment prediction and counseling, and for more objectively specifying the type of orthodontic treatment for CLP patients. Other sources of information-such as evaluation of psychological and socioeconomic status, and patient's wishes-are still necessary for finalizing the treatment plan.*

Keywords: *Cleft lip-palate, Orthognathic surgery prediction*

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A deficient maxilla is one of the most common problems in patients with cleft lip and/or palate (CLP), producing the characteristic Skeletal Class III pattern[1-3]. Generally, if the skeletal discrepancy is mild and esthetic concerns are minimal, dental compensation with orthodontic treatment alone may effectively camouflage the skeletal discrepancy[4]. However, for patients with a severe skeletal discrepancy, additional orthognathic surgery is required to position the maxilla and mandible for both acceptable esthetics and function. Attempted orthodontic compensation for the skeletal discrepancy, when orthognathic surgery is a later possibility, produces not only unsatisfactory esthetic results but also increases treatment time and reduces benefits versus costs. Such revisionary treatment is unlikely to be stable and may compromise periodontal health of the anterior teeth[5,6].

With the intention of trying to avoid such adverse outcomes, Singhawannakul[7] developed the Formula for Orthodontics and Surgery Prediction (FOSP)-an equation for predicting the need for additional orthognathic surgery or orthodontic treatment alone in CLP patients. The algorithm was developed using discriminate analysis of 59 cephalometric hard and soft tissue profile measurements (*viz.*, 25 skeletal, 13 dental and 23 soft tissue profile variables) from 119 subjects with different types of CLP treated at the Orthodontic Department, Faculty of Dentistry, Khon Kaen University. To prepare the analysis, an expert clinician, employing a general viewing of the clinical records of the 119 subjects, classified each subject into one of two groups of treatments-either orthodontics alone or orthognathic surgery.

The expected benefits *vs.* costs of what could be expected from the alternative treatments were taken into account by the expert. Based on the discriminate

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analysis, three cephalometric variables were found to incorporate into the FOSP. These factors were (a) the skeletal relationship (ANB angle) (b) upper incisor position (U1-APog in millimeters) and (c) lower lip profile relationship (L lip-N perpendicular line in millimeters). The FOSP was to be applied to all types of CLP without regard to age or sex.

The present study aimed to test the predictive value of the algorithm in determining the likely treatment plan, whether orthodontic treatment alone or orthognathic surgery, for CLP patients with Class III malocclusions and varying degrees of skeletal Class III deformity.

Material and Method

Following approval by the Ethics Committee of Khon Kaen University, the authors collected the pre-treatment lateral cephalometric films of 105 non-syndromic CLP patients with Class III malocclusions, without regard to age, sex or cleft type, from the Department of Orthodontics, Faculty of Dentistry, Khon Kaen University, Thailand. Subjects were included only if their final treatment with orthodontics alone or with orthodontics combined with orthognathic surgery was completed or in the process of completion. Their lateral cephalographs required good quality in sharpness, brightness and contrast. The present study excluded patients missing upper incisors or having a history of trauma affecting craniofacial growth and development.

Pre-orthodontic treatment cephalometric values of the ANB angle, U1-APog (mm) and L lip-N perpendicular line (mm) were measured and placed in the FOSP, taking into account positive or negative values. Repeat cephalometric tracing and measurements were made by an experienced examiner. Intra-examiner reliability was assessed using the intra-class correlation coefficient (ICC) Model 3 with SPSS® Version 13.0 (Statistical Package for Social Sciences for Windows). The discriminate scores (D) were then calculated using the FOSP[7]:

D = -1.861 - 0.378 ANB - 0.245 U1-APog + 0.129 L lip-N perpendicular line

Using the Receiver Operating Characteristic (ROC) curve, the critical D-score that represented an appropriate cut-off point in distinguishing the type of treatment was selected. All of the subjects were then separated into two groups-orthodontic treatment alone or additional orthognathic surgery-according to their respective calculated D-scores. The predictive value of each respective treatment plan derived from the FOSP was determined, using the actually received treatment as a reference. The actually received treatment plan of each subject was determined by the subject's attending orthodontist, with > 10 years' experience in treating CLP deformities.

Results

General characteristics of study subjects

The mean age of subjects, at the time that the pre-treatment lateral cephalographs were taken, was 12.97 $\pm$ 4.46 years (range, 5 to 29) with 71.4% being under 15 years of age (Table 1). Various cleft types were included. The number of subjects with unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), cleft lip with or without cleft alveolus (CL $\pm$ A) and cleft palate only (CP) are presented in Table 2. Actual received treatment categories, classified by sex, are shown in Table 3. The percentage of subjects who received orthodontic treatment combined with orthognathic surgery was ~45%, corresponding closely to the 46.2% predicted by Singhawannakul[7]. Approximately 20% of male subjects with UCLP received orthodontic treatment combined with orthognathic surgery, which is comparable to the 25% reported in several other studies[8-10].

Intra-reliability Test

Calculation of the intra-examiner reliability

Table 1. Age range classified by sex

Age Range (Years)	Sex		Total n (%)
	Male n (%)	Female n (%)	
5-11	18 (17.14)	26 (24.76)	44 (41.90)
12-14	15 (14.29)	16 (15.24)	31 (29.53)
15-18	9 (8.57)	12 (11.43)	21 (20.00)
19-29	4 (3.81)	5 (4.76)	9 (8.57)
Total	46 (43.81)	59 (56.19)	105 (100.00)
Mean (SD)	12.88 (4.46)	13.09 (4.51)	12.97 (4.46)

Table 2. Cleft type classified by sex

Cleft Type	Sex		
	Male n (%)	Female n (%)	Total n (%)
UCLP	24 (22.86)	37 (35.24)	61 (58.10)
BCLP	14 (13.33)	9 (8.57)	23 (21.90)
CL $\pm$ A	7 (6.67)	12 (11.43)	19 (18.10)
CP	1 (0.95)	1 (0.95)	2 (1.90)
Total	46 (43.81)	59 (56.19)	105 (100.00)

indicated excellent reproducibility of repeated cephalometric measurements. ICC values of the ANB angle, U1-APog (mm) and L lip-N perpendicular line (mm) are 0.998, 0.998 and 0.999, respectively.

Predictive quality of the FOSP formula

In reference to the ROC curve, the area under the curve was 0.796 (p-value < 0.001). The critical D-score of the FOSP in the present study was 0.648, yielding the greatest percentages for both sensitivity and specificity. A higher D-score indicated the need for orthognathic surgery; while a D-score below 0.648 signified orthodontic treatment alone. Substituting the three measurements for each subject in the FOSP calculation resulted in correct predictions for 79 subjects (75.2%) (Table 4). The respective sensitivity and specificity of the FOSP for prediction of the need for additional orthognathic surgery were 68.1% (52.9%-80.9%, 95%CI) and 81% (68.6%-90.1%, 95%CI). The high specificity indicated that the FOSP enabled identification of candidates for additional orthognathic surgery which would subsequently reduce the number of subjects misclassified as needing the surgery. The means and standard deviations (SD) of the D-scores, according to types of classification from the FOSP, are presented in Table 5.

Discussion

Until now, the establishment of objective criteria for determining the need for orthognathic surgery among Class III malocclusion patients with clefts has not been possible because of the range of factors influencing treatment decisions[10,11]. Several previous studies used the cephalometric analysis and prediction tracings to establish a model for predicting whether a patient could be treated by orthodontics alone or by combined orthodontics and orthognathic surgery[12-15]. However, for clinical application, variation among different populations must be taken into account. Being limited to lateral cephalometrics is, therefore, problematic, as the metrics represent only two dimensions of dento-skeletal traits.

Development of the FOSP represents an attempt to provide clinicians with a tool to perform early a quantitative and objective assessment for the optimal outcome; whether (a) definitive camouflage orthodontic treatment should proceed or (b) orthognathic surgery is necessary. The percentage of correctly classified subjects (accuracy) using the FOSP algorithm was 75.2%, which was less than the 83.2% estimated in the initial development of the FOSP, but which lacked the inclusion of treatment results which the present study made a point of including[7].

The prediction models of Nollet et al for cephalometric outcomes at age 18 were developed

Table 3. Actually received treatment categories classified by Sex

Treatment Categories	Sex		Total n (%)
	Male n (%)	Female n (%)	
Surgery	22 (46.81)	25 (53.19)	47 (44.76)
Non-surgery	24 (41.38)	34 (58.62)	58 (55.24)
Total	46 (43.81)	59 (56.19)	105 (100.00)

Table 4. Prediction results of the FOSP application

Predicted Treatment Plan	Actually Received Treatment Plan		Total n (%)
	Surgery n (%)	Non-surgery n (%)	
Surgery	32 (30.48%)	11 (10.48%)	43 (40.95%)
Non-surgery	15 (14.29%)	47 (44.76%)	62 (59.05%)
Total	47 (44.76%)	58 (55.24%)	105 (100.00%)

Table 5. Means, standard deviations (SD), minimum and maximum values of the D scores according to types of treatment classification from the FOSP

Classification	D score		
	Mean $\pm$ SD	Min	Max
Correctly classified as surgery group (n = 32)	3.00 $\pm$ 1.71	0.65	7.43
Correctly classified as non-surgery group (n = 47)	- 1.70 $\pm$ 1.50	- 5.77	0.56
Incorrectly classified as surgery group (n = 11)	1.93 $\pm$ 0.92	0.69	3.67
Incorrectly classified as non-surgery group (n = 15)	- 1.20 $\pm$ 1.30	- 4.48	- 0.01

from the cephalometric values taken at age 9 (in 40 UCLP subjects)[15]. They found four cephalometric measurements from among 14 angular and 2 ratios of hard and soft tissue traits (six skeletal, two dental and eight soft tissue profile variables) to be the most relevant cephalometric values for surgical prediction. These were SNA, SNPog, soft tissue ANB and soft tissue ANPog. The actual need for orthognathic surgery in the study was determined by a panel of two orthodontists and one maxillofacial surgeon. Their recommended treatment-need compared with the study's identified need for surgery at adulthood was correctly predicted with 85% accuracy, which is higher than the predictive value of the FOSP in our study. However, the number of subjects in their study, especially in the surgical group (11 subjects), was limited.

Differences in study population traits must be considered when attempting to explain differences in the predictive values between the studies. The lower percentage of predictive accuracy in the presented study-compared with the studies of Singhawannakul and Nollet et al-may be in part due to those studies not having been compared with the actual treatment accepted by each patient, which was one of the pre-conditions of the present study. The predictive merit of the FOSP in the present study by Singhawannakul is questionable since it was based on the predictive capabilities of only one expert clinician. Moreover, the findings of that study were diluted by the large variations in the age and cleft condition of the subjects included. It must also be noted that the present study used the same sample population as was used in the previous development of the FOSP, so that the results may not be transferable to other populations with different ethnic and socio-economic backgrounds.

The accuracy of the present study was also less than the 92.0% of the treatment prediction equation developed by Stellzig-Eisenhauer et al[16]; however, their research was based on adult Class III non-cleft subjects, with no possibility of favorable jaw growth, while 71.4% of the subjects in the present study were under 15 with incomplete jaw growth. The existence of cleft deformities may variously affect later development of the subjects, thereby increasing the difficulty in predicting the later treatment that would be needed. The interval between timing of treatment prediction and the definitive treatment plan decision-which is usually done at between 15 and 18 years of age-might also play a role in the accuracy of treatment prediction. The longer the interval between the time the treatment is predicted and when it is given, the more likely it is that there will be a discrepancy between them.

In the misclassified groups, the mean D-scores calculated from the FOSP were closer to those of the opposite group of actual treatment needed (Table 5). This suggests that using only the three cephalometric values of the FOSP was insufficiently sensitive to accurately predict the type of orthodontic treatment needed, especially for borderline surgical cases. Although the FOSP was developed from many more variables for both hard and soft tissue profiles (*i.e.* 60 cephalometric measurements, compared to only 16 variables in the present study by Nollet et al), it is important to keep in mind that there may be other factors essential in treatment planning. These would include maxillary midline discrepancies, facial symmetry, dentofacial esthetics, dento-alveolar health, likely long-term stability, comparative treatment costs and risks, and other non-measurable factors such as patient's concerns about pain, discomfort, and hospitalization involved with a surgical procedure[6,10,16,17]. Such factors are difficult to assess without communication with the patient because the patient's decision may contradict the clinician's recommendation.

Conclusion

Using only three cephalometric measurement values for the final treatment decision may be insufficient as it relies on multiple sources. Currently, the FOSP is the most useful tool for advising parents of possible treatment, for the purpose of avoiding the clinical and psychological problems arising from orthodontic camouflage for patients who later are dissatisfied with the treatment-outcome and request corrective surgery. Early prediction of the final treatment need also provides an opportunity to identify the future service needs at the cleft care center, thereby aiding the manpower assessment for orthodontists and maxillofacial surgeons. Further evaluation of the effect of age, sex and cleft type on the predictive value of the FOSP for treatment prediction is recommended.

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Potential conflicts of interest

None.

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ความสามารถในการพยากรณ์ด้วยสมการแนะนำแผนการรักษาทางทันตกรรมจัดฟันในผู้ป่วยปากแหว่งเพดานโหว่ที่มีการสบฟันผิดปกติชนิดที่ 3

ทัศนีย์ วังศรีมงคล, มนเทียร มโนสุดประสิทธิ์, คีท ก็อดฟรีย์, ชุตินารถ คูรัตน์ชัชวาล, พลากร สุรกุลประภา

วัตถุประสงค์: *เพื่อประเมินประสิทธิภาพของแผนการรักษาที่ได้จากสมการพยากรณ์แผนการรักษาทางทันตกรรมจัดฟัน (FOSP)*

วัสดุและวิธีการ: *ทำการรวบรวมข้อมูลจากประวัติการรักษาทางทันตกรรมจัดฟันของผู้ป่วยปากแหว่งเพดานโหว่ชนิดไม่มีกลุ่มอาการร่วมที่มีอายุระหว่าง 5-29 ปี และมีการสบฟันผิดปกติชนิดที่ 3 ที่ระดับความรุนแรงต่าง ๆ จำนวน 105 ราย บันทึกค่าจากภาพรังสีกะโหลกศีรษะด้านข้างในระยะก่อนได้รับการรักษาทางทันตกรรมจัดฟันจำนวน 3 ค่า เพื่อเข้าสมการ FOSP โดยใช้เส้นโค้งอาร์โอซีช่วยกำหนดค่าคะแนนจุดตัดที่เหมาะสมในการใช้จำแนกแผนการรักษาด้วยการพยากรณ์ในผู้ป่วยแต่ละรายว่าสามารถรักษาด้วยการจัดฟันเพียงอย่างเดียว หรือร่วมกับการผ่าตัดขากรรไกร จากนั้นทำการเปรียบเทียบระหว่างแผนการรักษาที่ได้จากสมการกับแผนการรักษาจริงที่ได้รับ*

ผลการศึกษา: *ที่คะแนนจำแนกจุดตัดที่ 0.6475 แผนการรักษาที่พยากรณ์ได้จากสมการ FOSP สามารถพยากรณ์ได้ถูกต้องในผู้ป่วยจำนวน 79 ราย (75.2%) โดยมีค่าความไวและความจำเพาะของสมการ FOSP ในการพยากรณ์แผนการรักษาร่วมกับการผ่าตัดขากรรไกรที่ 68.1% (52.9%-80.9%) และ 81.0% (68.6%-90.1%) ตามลำดับ*

สรุป: *สมการ FOSP ช่วยในการพยากรณ์แผนการรักษาในระยะแรก และเป็นแนวทางในการให้คำแนะนำแผนการรักษาทางทันตกรรมจัดฟันในผู้ป่วยปากแหว่งเพดานโหว่ได้อย่างเป็นรูปธรรม อย่างไรก็ตามในการตัดสินใจให้การรักษาในระยะสุดท้ายยังคงต้องอาศัยข้อมูลจากแหล่งต่างๆ ซึ่งรวมถึงสภาวะทางจิตใจ เศรษฐานะ และความต้องการของผู้ป่วย*