



Title	Predicting Need for Orthognathic Surgery in Children with Cleft Lip and/or Palate
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論文内容の要旨

氏 名 (林 在娟)	
論文題名	Predicting Need for Orthognathic Surgery in Children with Cleft Lip and/or Palate (口唇口蓋裂を伴うこどもにおける外科的矯正治療の必要性の予測)
論文内容の要旨	
Introduction Determining risk factors and predicting prognosis for orthognathic surgery (OGS) for children with cleft lip and/or palate (CL/P) are crucial not only for reducing risks of deterioration but also for developing effective treatment plans in early stage to reduce clinical burdens. While numbers of risk factors have been reported, these were hardly considered simultaneously. Recently, machine-learning methods are available for predicting prognosis examining multiple variables comprehensively. This study aimed at developing comprehensive mathematical models to determine the risks and predict the need for OGS in mixed dentition patients with CL/P by using lateral cephalograms, cleft type, severity of lip separation at birth, the number of missing teeth, sex, surgery methods of palatal closure and surgeons, duration of orthodontic treatment (Experiment 1), dental morphology from three-dimensional (3D) digital models (Experiment 2). Methods Experiment 1. In this retrospective cohort study, we included 126 patients with bilateral and unilateral CL/P (70 males, 56 females; 19 BCLP, 5 BCLA, 71 UCLP, 20 UCLA, 11 CP) who visited Orthodontic Department, Osaka University Dental Hospital between 2002 and 2014 and took at least three lateral cephalograms at 7 (T1), 10 (T2), and 15 (T3) years old. As predictor variables, type of cleft, severity of lip separation at birth (absence of Simonart's band), the number of missing teeth, sex, surgery methods of palatal closure (1-step Push-back or 2-step Furlow's method) and surgeons, lateral cephalograms at T1 and T2, growth between T1 and T2, and total duration of orthodontic treatment (maxillary protraction and expansion) were examined; and as outcome variables, clinical decision of the need for OGS, skeletal discrepancy ($ANB \geq 0$ or $< 0^\circ$), and overjet (≥ 0 or $< 0\text{mm}$) at T3 were examined. Combinations of the predictor variables observed at T1 and T2 were entered into multivariable logistic regression models to predict the outcomes ($p=0.05$). Prediction power was evaluated with the area under the receiver operating characteristic curve (AUC). Experiment 2. Among 126 patients in Experiment 1, 80 patients who had dental plaster models at T1 were enrolled. Fifty-six patients had clinical decision of the need for OGS (OGS group) and 24 were camouflage cases (non-OGS group) at T3. Plaster models at T1 were scanned with three-dimensional (3D) scanner (3Shape Dental System, 3Shape Co., Copenhagen). To compare transverse and palatal features between OGS and non-OGS groups, a total of 32 landmarks (24 maxillary, 8 mandibular) were digitized on the 3D models with HBM Rugle (Medic Engineering Co., Kyoto). The coordinate values were then analyzed through principle component (PC) analysis and mean shape comparisons using geometric morphometrics software (MorphoJ 1.07a, Klingenberg). Unilateral clefts on the right side were inverted to the left side to examine the effect of cleft. Further, to examine if the prediction accuracy can be improved by using variables extracted from 3D models, two multivariable logistic regression models with and without aforementioned PCs were developed to predict the clinical decision of the need for OGS. Prediction power was evaluated with the AUC.	

Results

Experiment 1. A total of six mathematical models was developed and the mean AUC was 0.93 (0.91–0.99) indicating high predicting accuracy (AUC >0.9). Determined risk factors were cleft type (BCL/P), Push-back method of palatal closure, male, smaller protrusion and length of maxillary, smaller length of anterior cranial base, greater mandibular protrusion, greater increase of lower facial height, smaller clockwise rotation of palatal and mandibular plane between T1 and T2, the number of missing teeth, steeper palatal and mandibular plane at T1, T2. The total duration of orthodontic treatment, surgeons of palatal closure, severity of lip separation at birth showed no significant relationships for the prognosis.

Experiment 2. The PC analysis resulted in two components explaining 20.0% and 14.5% of the total shape variance. Dental models of OGS group showed several distinctive features compared to non-OGS group; shallow palatal, palatally positioned and inclined upper C on the cleft side, palatally positioned and buccally inclined (dentally compensated) upper E, buccally positioned and lingually inclined (dentally compensated) lower E, crossbite tendency on E and cleft-side C, and anteriorly positioned lower C. PC1 indicated shallow and narrow maxilla. The AUCs for the prediction models with and without PCs showed 0.80 and 0.75 respectively, indicating morphological features of the 3D dental models improved prediction accuracy for the clinical decision of the need for OGS.

Conclusions

Comprehensive mathematical models to predict the need for OGS in mixed dentition patients with CL/P were developed indicating high predicting accuracies. Determined risk factors were cleft type (BCL/P), Push-back method of palatal closure, male, smaller protrusion and length of maxillary, smaller length of anterior cranial base, greater mandibular protrusion, greater increase of lower facial height, smaller clockwise rotation of palatal and mandibular plane between 7 and 10 years old, the number of missing teeth, steeper palatal and mandibular plane at 7 and 10 years old. Shallow and narrow maxilla in 3D dental models at 7 years old were also found to be a predictor for the clinical decision of the need for OGS at 15 years old.

論文審査の結果の要旨及び担当者

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論文審査の結果の要旨 口唇口蓋裂を有する混合歯列期の患者において、将来的な外科的矯正治療の必要性を予測しそのリスク因子を同定することは、最適な治療計画の立案や患者の治療負担を軽減するために重要である。本研究は、多変量ロジスティック解析を用いて口唇口蓋裂を有する患者における将来的な予後のリスク因子を同定したものであり、博士（歯学）の学位論文として価値のあるものと認める。			