Journal of Plastic, Reconstructive & Aesthetic Surgery (2019) 72, 1813–1818

JPRAS
An International Journal of Surgical Reconstruction
www.JPRASurg.com

Orthodontic premaxillary setback versus premaxillary osteotomy with gingivoperiosteoplasty for Bilateral cleft lip and palate patients: 4-year observation outcomes

Shinji Kobayashi[a,*], **Takashi Hirakawa**[b], **Yasunari Sasaki**[c], **Toshihiko Fukawa**[d], **Jiro Maegawa**[e]

[a] *Department of Plastic and Reconstructive Surgery, Kanagawa Children's Medical Center*
[b] *Hirakawa Orthodontic Clinic*
[c] *Department of dentistry, Kanagawa Children's Medical Center*
[d] *Fukawa Orthodontic Office*
[e] *Department of Plastic and Reconstructive Surgery, Yokohama City University Hospital*

Received 4 May 2019; accepted 24 May 2019

KEYWORDS
Orthodontic premaxillary setback;
Premaxillary osteotomy;
Gingivoperiosteoplasty;
Bilateral cleft lip and palate;
Furlow double-opposing Z palatoplasty

Summary Patients with bilateral cleft lip and palate (BCLP) generally require several stages of treatment, and adequate maxillary development without orthognathic surgery is not easy to achieve. The purpose of this study was to determine short-term outcomes of orthodontic premaxillary setback (OPS) and premaxillary osteotomy (PO) for complete BCLP treatment.
Patients and Methods: Twenty-six patients with BCLP were consecutively treated for 4 years by a single surgeon. All patients were categorized by three indications for the first operation: OPS1, cheiloplasty + gingivoperiosteoplasty (GPP) + palatoplasty; OPS2, cheiloplasty + GPP; and PO, cheiloplasty + GPP + PO. Cephalograms for maxillary growth and velopharyngeal function (VPF) were judged at 4 years old, before orthodontic treatment.
Results: OPS1 was performed in 14 cases, OPS2 in 8 cases, and PO in 4 cases. As for cephalometric analysis at 4 years old, no significant differences between groups were seen in any cephalometric measurements. As for speech outcomes, assessment of VPF at 80.8±14.8 months was good in 17 cases, slightly impaired in 8 cases, and marginally impaired in 1 case, with no severely impaired cases.

* Address for correspondence: Shinji Kobayashi, Kanagawa Children's Medical Center, Department of Plastic and Reconstructive Surgery, Mutsukawa 2-138-4, Minami-ku, Yokohama, Kanagawa, Japan.
E-mail address: skobayashi@kcmc.jp (S. Kobayashi).

https://doi.org/10.1016/j.bjps.2019.05.047

Discussion: No significant differences in maxillary growth or speech outcomes were seen between OPS1, OPS2, and PO groups at 4 years old, possibly because all groups showed the same position of the premaxilla after the first operation.
Conclusions: No significant differences in maxillary growth or speech outcomes were seen for 26 patients with BCLP between OPS1, OPS2, and PO groups at 4 years old. However, the long-term growth characteristics remain unclear.

Background

Patients with bilateral cleft lip and palate (BCLP) generally require several stages of treatment. Adequate maxillary development without orthognathic surgery is difficult to achieve. Patients with BCLP undergo many operations by adulthood, and the frequency of orthognathic surgery is greater among patients with BCLP than among patients with unilateral cleft lip and palate.[1,2]

Further, orthodontic premaxillary setback (OPS) in the form of nasoalveolar molding and gingivoperiosteoplasty (GPP) has been expected to not only improve the shape of the lip and nose but also obviate the need for alveolar bone graft (ABG) by forming alveolar bone,[3-9] although some studies have reported concerns over maxillary growth retardation and lack of long-term benefits.[10-16]

However, if OPS was not performed or was unsuccessful, the facial appearance would be left until premaxillary osteotomy (PO) is performed for BCLP with a protruded premaxilla. Some studies have reported patients with BCLP with a protruded premaxilla who underwent PO from approximately 8 to 12 years old.[17-22] Patients with BCLP will need to spend time with a protruded face in kindergarten and elementary school if premaxillary setback such as PSO and PO is not performed as early as possible.

We have performed OPS and PO as the primary operations to prevent patients from spending more time with a protruded face. The purpose of this study was to determine short-term outcomes of OPS and PO for complete BCLP treatment.

Methods

Twenty-six consecutive patients with BCLP without mental disorders and serious anomalies were treated for 4 years since 2008 by a single surgeon. All patients registered at Kanagawa Children's Medical Center were retrospectively reviewed. Ethics approval was granted by our institution, and informed consent was obtained from surrogates (approval number 107-10).

First, OPS was performed in all patients to set back the premaxilla. Specifically, OPS was aimed at achieving a distance of 0 mm between the anterior line of the lateral segment and the posterior line of the premaxilla (DAP) (Figure 1). In cases where the premaxilla could not be sufficiently set back by OPS, primary PO with cheiloplasty was performed to achieve setback as the first operation. In addition, cleft width (CW) was measured before and after OPS.

All patients were categorized retrospectively by three indications: 1) cheiloplasty, GPP, and Furlow double-opposing Z palatoplasty (FDOP) performed simultaneously (OPS1 group); 2) cheiloplasty and GPP performed simultaneously, followed by FDOP (OPS2 group); and 3) cheiloplasty, PO, and GPP performed simultaneously, followed by FDOP (PO group).

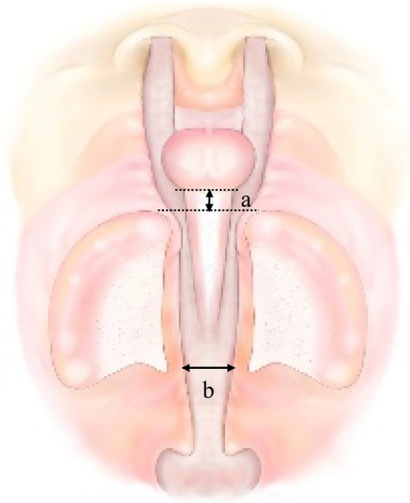

Figure 1 Measurement of the distance between the anterior line of the lateral segment and the posterior line of the premaxilla (DAP) and cleft width (CW). a) DAP; b) CW.

In all patients at 4 years of age before orthodontic treatment, a cephalogram was taken to analyze maxillary growth. Velopharyngeal function (VPF) was judged using the Japanese Scale for Assessment of Cleft Palate Speech on a 4-point scale: good, slightly impaired, marginally impaired, or severely impaired. This battery consisted of speech evaluation (Japanese vowels, single syllables, 50 words, and several sentences, as well as conversation) combined with the volume of nasal air escape during soft blowing. Depending on an assessment of hypernasality and consonant distortion due to nasal emission, each case was judged on a 4-point scale: normal, mild, moderate, and severe.[23-25]

SPSS for Windows version 24.0 software (SPSS, Chicago, IL) was used for statistical analysis. Data were compared between the three groups using the Kruskal–Wallis test, followed by the Mann–Whitney U test. Data are expressed as mean±standard deviation.

Results

OPS1 was performed for 14 patients (53.8%), OPS2 for 8 patients (30.8%), and PO for 4 patients (15.4%). For OPS,

Table 1 Beginning and ending times, DAP and CW measurements before and after OPS.

Group	The beginning /the ending time of PSO(days)(mean±SD)	DAP before/ after PSO(mm)	CW before/after PSO(mm)
OPS1	13.8 ± 11.8/211.2 ± 35.4	2.7 ± 3.6/0 ± 0	13.3 ± 2.6/8.7 ± 1.3**
OPS2	9.1 ± 10.8/235.4 ± 42.8	4.3 ± 2.7/0 ± 0	14.6 ± 2.0/12.4 ± 1.7
PO	41±42.6/205.0 ± 7.4	5.1 ± 3.7/0.8 ± 1.5	14.3 ± 0.6/10.5 ± 1.9
Total	16.8 ± 20.9/ 217.2 ± 35.8	3.4 ± 3.3/0.1 ± 0.6	13.8 ± 2.2/10.2 ± 2.3

(*P<.01, **P<.05).

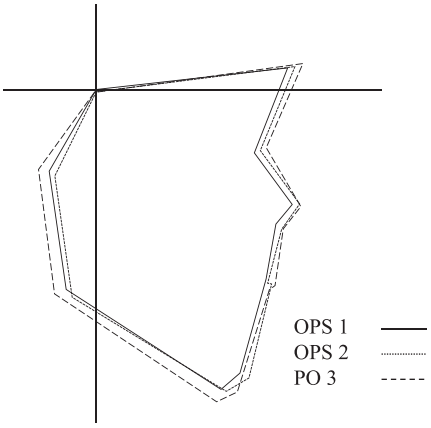

Figure 2 Cephalometric analysis at 4 years old. Group 1: solid line; Group 2: dotted line; Group 3: dashed line.

Table 2 Speech outcomes.

speech outcomes	Total	OPS1	OPS2	PO
No.	26	14	8	4
VPF				
good	17	11	3	3
slightly impaired	8	3	4	1
marginally impaired	1	0	1	0
severely impaired	0	0	0	0
hypernasality				
normal	18	11	4	3
mild	8	3	4	1
moderate	0	0	0	0
severe	0	0	0	0
consonant distortion				
normal	19	11	4	4
mild	7	3	4	0
moderate	0	0	0	0
severe	0	0	0	0

beginning and end times were 13.8±11.8 and 211.2±35.4 days in OPS1, respectively. DAPs before and after OPS were 2.7±3.6 and 0±0 mm, respectively. CWs before and after OPS were 13.3±2.6 and 8.7±1.3 mm, respectively. In OPS2, beginning and end times were 9.1±10.8 and 235.4±42.8 days, respectively. DAPs before and after OPS were 4.3±2.7 and 0±0 mm, respectively. CWs before and after OPS were 14.6±2.0 and 12.4±1.7 mm, respectively. In Group PO, beginning and end times were 41±42.6 and 205.0±7.4 days, respectively. DAPs before and after PSO were 5.1±3.7 and 0.8±1.5 mm, respectively. CWs before and after OPS were 14.3±0.6 and 10.5±1.9 mm, respectively. CW after OPS was significantly lower in OPS1 than in OPS2 and PO (P<.01). No significant differences (P>.05) were seen between groups for other items (Table 1). As for cephalometric analysis at 4 years old, no significant intergroup differences were seen in any cephalometric measurements (P>.05) (Figure 2, supplementary 1).

As for speech outcomes at a mean age of 80.8±14.8 months (range, 4 years 0 months to 9 years 9 months), VPF was assessed as good in 17 cases (65.4%), slightly impaired in 8 cases (30.8%), and marginally impaired in 1 case (3.8%), with no severely impaired cases. Hypernasality was assessed as normal in 18 cases (69.2%) and mild in 8 cases (30.8%), with no moderate or severe cases. Assessment of consonant distortion was normal in 19 cases (73.1%) and mild in 7 cases (26.9%), with no moderate or severe cases. Speech outcomes tended to be better for OPS1 cases than for OPS2 cases (Table 2).

Clinical Reports

Representative Case from OPS1: A boy with BCLP with protruded and deviated premaxilla

A markedly protruded and deviated premaxilla was repositioned by OPS at 2 months old. At 6 months old, bilateral simultaneous cheiloplasty, GPP, and FDOP were performed simultaneously. At 4 years old, occlusion was good, and computed tomography (CT) at 5 years old revealed good bone formation on both alveoli, meaning that corrective treatment was possible without ABG (Figure 3, supplementary 2 and 3).

Representative Case from OPS2: A boy with BCLP with protruded premaxilla and width cleft

The markedly protruded premaxilla was repositioned by OPS at 5 months old. At 8 months old, bilateral simultaneous cheiloplasty and GPP were performed together, followed by palatoplasty at 1 year and 4 months old. At 4 years old, molar occlusion was edge-to-edge. CT at 5 years old showed insufficient bone formation on the right alveolus. ABG was performed subsequently on the right side (Figure 4, supplementary 4 and 5).

Representative Case from PO: A boy with BCLP with protruded premaxilla

The markedly protruded premaxilla was repositioned by OPS at 5 months old. At 8 months old, bilateral simultaneous cheiloplasty, PO, and GPP were performed together, followed by palatoplasty at 1 year and 4 months old. At 4 years old, molar occlusion was edge-to-edge. CT at 5 years old

Figure 3 Group 1: A boy with BCLP.
Left above: Preoperative appearance before OPS.
Left below: Preoperative intraoral findings after OPS.
Right: After one-stage operation, appearance and intraoral findings at 6 years old.

Figure 4 Group 2: A boy with BCLP.
Left above: Preoperative appearance before OPS.
Left below: Preoperative intraoral findings after OPS.
Right: After two operations, appearance and intraoral findings at 5 years old.

showed insufficient bone formation on the right alveolus. ABG was subsequently performed on the right side (Figure 5, supplementary 6 and 7).

Discussion

We showed short-term outcomes of OPS and PO for patients who underwent complete BCLP treatment during 4 years. As a result, no significant differences in maxillary growth or speech outcomes were seen between OPS groups (OPS1 and OPS2) and PO group at 4 years old. This might have been because all groups showed the same position of the premaxilla (DAP = 0 mm) after the first operation. If the position of the premaxilla was set back further, maxillary growth might have been worse.

Figure 5 Group 3: A boy with BCLP. Preoperative appearance before OPS.
Left above: Preoperative appearance before OPS.
Left below: Preoperative intraoral findings after OPS.
Right: After two operations including cheiloplasty, GPP and PO, and palatoplasty; appearance; and intraoral findings at 5 years old.

In this study, we performed estimations at 4 years old to evaluate natural maxillary growth because we have used orthodontic treatment with a maxillary protraction appliance[26] after 4 years old for patients showing anterior cross-bite. In the future, the premaxilla might retreat further due to lip pressure after cheiloplasty and synostosis at the suture,[27] resulting in anterior cross-bite. We need longer follow-up and accumulation of more cases, although a certain level of maxillary growth retardation might not be unavoidable due to numerous surgeries and original underdevelopment of the lateral maxillary segments in the embryonic period.[28]

Early OPS or PO proved to be highly useful in terms of improving nasal and lip shape, including elongation of the columella and nasolabial angle, as well as occlusion. These methods allow the child to fit into society when attending kindergarten and elementary school[29] because the facial structures are improved from the first operation. Some studies have reported on patients with BCLP with a protruded premaxilla who underwent PO with ABG from approximately 8 to 12 years old.[17-22] In such cases, most patients must spend time with a protruded face in kindergarten and elementary school. Our strategy was considered to significantly lower parental stress because the strategy was to ensure repair of the premaxilla at the first operation.[30-32]

Furthermore, FDOP could be performed simultaneously with cheiloplasty in 15 cases (55.6%) in which the CW was less than 10 mm. This strategy was helpful for reducing the number of operations and obtaining good speech outcomes because speech could be learned earlier.

The present report described only a small number of cases with relatively short follow-up, but maxillary growth and speech outcomes were acceptable. Careful follow-up is

needed until the patient reaches adulthood, especially for maxillary growth.

Conclusions

For 26 patients with complete BCLP treatment, no significant intergroup differences in cephalometric measurements were seen between OPS and PO groups, and no cases showed severe impairment in terms of VPF. However, the characteristics of long-term growth remain unclear.

Acknowledgment

No support was received.

Funding

None.

Conflicts of interest

None declared.

Ethical approval

Not required.

Supplementary material

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.bjps.2019.05.047.

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