Original Article

Root development differences between cleft-adjacent teeth on the cleft side in comparison to their analogs on the noncleft side in patients with nonsyndromic cleft lip and palate who received secondary alveolar bone grafting

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ABSTRACT

Objective: To assess differences in root development between the cleft side (CS) and noncleft side (NCS) for permanent maxillary central incisor and canine longitudinally in patients with nonsyndromic complete unilateral cleft lip and palate (cUCLP) who received secondary alveolar bone grafting (SABG) and to evaluate the effects of SABG on the acceleration of root development of these teeth.

Materials and Methods: Permanent maxillary central incisors and canines of 44 subjects with nonsyndromic cUCLP who had all their cleft-related surgeries performed by the same surgeon were analyzed retrospectively from chart notes and radiographs. Panoramic and periapical radiographs at time point 1 (T1) (age, 7.55 years), at SABG (time point 2 [T2], 10.13 years), and a minimum of 2 years after SABG were studied. Root development rating scores on the NCS and CS were compared using paired *t*-tests and analyses of proportions.

Results: Mean root development score differences (NCS − CS) for canines and central incisors were greatest at T2 but diminished at time point 3 (T3). A larger proportion of teeth on the CS trailed the teeth on the NCS by at least 1 point at T2 than at T1 or T3, with the smallest proportion being observed at T3. The change in root development scores from T1 to T2 and from T2 to T3 showed relative CS acceleration from T2 to T3, indicating a catch-up of root development of cleft-adjacent teeth after SABG.

Conclusions: Root development of cleft-adjacent central incisors and canines is slow in comparison with their noncleft analogs. Root development of these teeth accelerates following SABG. (*Angle Orthod*. 2024;94:75–82.)

KEY WORDS: Root development; UCLP; SABG; Cleft

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Table 1. Published Literature on the Influence of SABG on the Root Development of Permanent Maxillary Teeth in Patients with UCLP[a]

Authors	Sample(s)	Teeth Measured	Longitudinal Assessments	Findings
El Deeb et al.[6] 1982	28 UCLP and UCLA (combined sample)	Permanent maxillary canines	T1 (before SABG): no T2 (at SABG): yes T3 (post-SABG): yes, mean 4.3 ± 1.5 y post-SABG	No significant difference in root development of CS vs NCS canines at grafting or upon eruption. All canines completed root development.
Park et al.[10] 2014	25 UCLP and 25 UCLA	Permanent maxillary central incisors and canines	T1 (before SABG): no T2 (at SABG): yes T3 (post-SABG): yes, 1 y post-SABG	Central incisor root development was not different at SABG and 1 y after SABG between the CS and NCS. CS canines in UCLP are delayed at SABG vs the NCS but develop faster than the NCS after SABG.
Nishihara et al.[18] 2014	25 UCLP (n = 16 pre-canine eruption SABG, n = 9 post-canine eruption SABG)	CS permanent maxillary canines	T1 (before SABG): no T2 (at SABG): yes T3 (post-SABG): yes, 1 y and >4 y post-SABG	Root development advanced in SABG after canine eruption group vs SABG before canine eruption group at SABG and at 1 y post-SABG. No significant difference in root development between groups at >4 y post-SABG.
Vellone et al.[19] 2017	24 UCLP	Permanent maxillary canines	T1 (before SABG): yes, 1 y before SABG T2 (at SABG): no T3 (post-SABG): yes, 1 y post-SABG	No significant differences in CS canine root formation before and after SABG compared to the NCS.
Vandersluis et al.[20] 2020	44 UCLP (n = 21 pre-canine emergence SABG, n = 23 post-canine emergence SABG)	Permanent maxillary central incisors and canines	T1 (before SABG): yes (age, 7 y) T2 (at SABG): yes T3 (post-SABG): yes, minimum of 2 y after SABG	SABG did not seem to adversely affect root development. The root development of the central incisors was not significantly different between SABG groups or between the CS and NCS. Relative to the post-canine emergence SABG group, the CS permanent maxillary canine in the pre-canine emergence SABG group developed slowly prior to SABG but seemed to accelerate after SABG.
Kadi et al.[21] 2021	30 UCLP and UCLA (combined sample)	Permanent maxillary canines	T1 (before SABG): yes, 6 mo before SABG T2 (at SABG): no T3 (post-SABG): yes, 6 mo after SABG	Measured total tooth length. The canines were longer after SABG than before. No significant difference in canine length between the CS and NCS.
Desai et al.[22] 2021	30 unilateral or bilateral cleft lip, alveolus, and palate (combined sample)	CS permanent maxillary canines	T1 (before SABG): no T2 (at SABG): yes T3 (post-SABG): yes, 1, 3, and 6 mo after SABG	Statistically significant development of the CS canine root after SABG.

[a] UCLP indicates unilateral cleft lip and palate; UCLA, unilateral cleft lip and alveolus; SABG, secondary alveolar bone grafting; T1, time point 1; T2, time point 2; T3, time point 3; CS, cleft side; and NCS, noncleft side.

INTRODUCTION

An increased frequency of dental developmental anomalies has been reported in patients with cleft lip and palate (CLP), with these differences being more pronounced in the immediate cleft vicinity.[1] One specific dental alteration that has been reported in patients with CLP is a delay in root development, both relative to age-matched controls[2–5] and when comparing the cleft side (CS) to the contralateral noncleft side (NCS).[2,3,6–12] Patients with CLP are three to four times more likely to have a difference of at least one stage of root and/or crown development between the CS and the NCS in comparison to contralateral sides in control populations.[9,13,14]

The presence of a cleft of the maxillary alveolar ridge itself has also been found to inhibit normal dental

Table 2. Root Development Rating System Based on Relative Crown and Root Lengths (from El Deeb et al.[6])

Root Development Score	Description
0	Crown completely formed, no root formation
1	Initial root formation
2	1/4 root formation (root < crown)
3	1/2 root formation (root = crown)
4	3/4 root formation (root > crown)
5	Complete root formation with open apex
6	Complete root formation with closed apex

development.[15,16] In patients with complete unilateral cleft lip and palate (cUCLP), a missing lingual cortical plate in the cleft site has been hypothesized to delay root development due to the lack of space for the root to develop.[10,17] Secondary alveolar bone grafting (SABG) in the mixed dentition is a commonly performed surgical procedure to repair clefts of the maxillary alveolus and is usually undertaken prior to the eruption of the CS permanent maxillary canine. While the continuity of the alveolar ridge provided by SABG may have a positive effect on root development, surgical treatment may impede root development due to a decreased blood supply and scar tissue formation.[11]

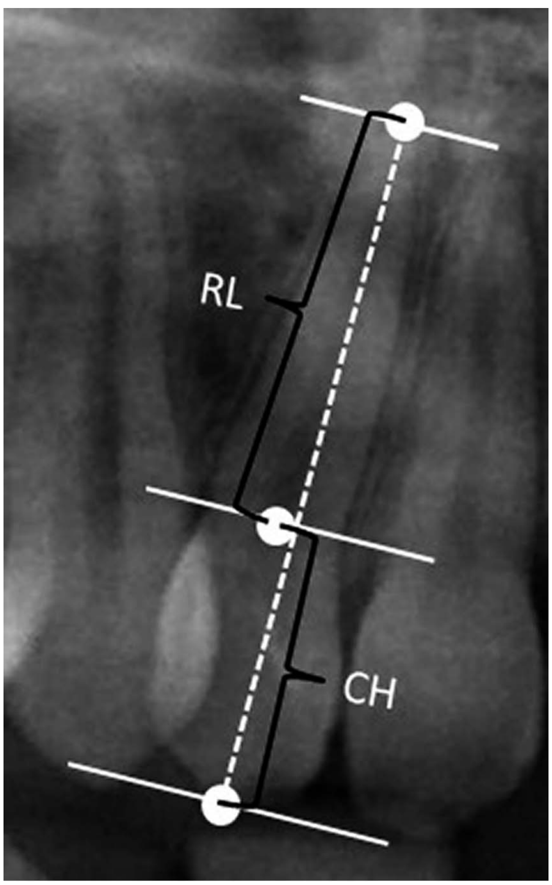

Figure 1. Crown height and root length measurements for root development rating scores. The long axis of the tooth (white dashed vertical line) and incisal, cervical, and apical lines (white solid horizontal lines) are drawn. Relative to tooth width, midpoints of the incisal, cervical, and apical lines are marked (white circles). Crown height (CH) is the vertical distance from the midpoint of the incisal line to the midpoint of the cervical line. Root length (RL) is the vertical distance from the midpoint of the apical line to the midpoint of the cervical line. CH and RL are compared to determine the root development rating score.

Table 3. Characteristics of the Included Sample of Patients[a]

Characteristic	Value
Male/female patients	27/17
cUCLP side, right/left	16/28
Age at T1, y	7.55 ± 0.90
Age at T2, y	10.13 ± 0.79
Age at T3, y	14.19 ± 1.63
Duration post-SABG at T3, y	4.02 ± 1.48

[a] cUCLP indicates complete unilateral cleft lip and palate; T1, time point 1; T2, time point 2; and T3, time point 3.

A few studies have investigated the influence of SABG on root development (Table 1).[6,10,18–22] An acceleration of root development of the permanent maxillary canine following SABG performed before canine eruption has been previously reported without any adverse effects on root development.[10,20] Other studies have also reported minimal adverse effects of SABG on tooth development, but these studies were limited by subjects not always being separated based on cleft severity,[6,21,22] surgical treatment being provided by more than one surgeon,[21] or a lack of clarity as to whether the included patients were treated by more than one surgeon.[6,10,18,19,22] As the experience of the surgeon can affect the outcomes of SABG,[23] and root development can be influenced by cleft severity,[13] these factors ideally should be controlled for. Additionally, while all studies measured root development at more than one time point, a longitudinal assessment of root development from a time prior to SABG, to the time of SABG, to a time after SABG has not been reported by most. Most studies also measured only the permanent maxillary canine[6,18,19,21,22] and not the maxillary central incisor (Table 1).

To better understand the longitudinal effects of SABG on the root development of cleft-adjacent teeth, the objectives of this study were (1) to determine the root development differences between cleft-adjacent teeth on the CS and their analogs on the NCS over time in patients with nonsyndromic cUCLP who received SABG by the same surgeon and (2) to explore the potential acceleration of root development following SABG.

MATERIALS AND METHODS

A retrospective longitudinal assessment of root development in patients with nonsyndromic cUCLP was performed at The Hospital for Sick Children in

Table 4. Comparison of Mean CS and NCS Root Development Scores of the Maxillary Central Incisors and Canines at the Three Time Points[a]

	Root Development Score					
	NCS		CS			
	Mean	SD	Mean	SD	Mean Difference (NCS − CS)	Paired *t*-Test *P* Value
Central Incisor						
T1 (7.55 ± 0.90 y)	3.31	1.22	3.26	1.11	0.05	1
T2 (10.13 ± 0.79 y)	5.45	0.69	5.1	0.87	0.35	.01*
T3 (14.19 ± 1.63 y)	6	0	5.93	0.26	0.07	.16
Canine						
T1 (7.55 ± 0.90 y)	1.8	0.79	1.56	0.76	0.24	.04*
T2 (10.13 ± 0.79 y)	3.69	0.89	3.28	0.91	0.41	.02*
T3 (14.19 ± 1.63 y)	5.87	0.5	5.58	0.8	0.29	.01*

[a] NCS indicates noncleft side; CS, cleft side; SD, standard deviation; T1, time point 1; T2, time point 2; and T3, time point 3.

* Statistically significant, $P < .05$.

Toronto, Canada. The study design was reviewed and approved by the research ethics board of The Hospital for Sick Children and the University of Toronto prior to study initiation. The management of patients with cUCLP from birth up to and including SABG surgery followed the same standardized protocol. All subjects had been treated with infant orthopedics, initiated soon after birth until primary cheiloplasty, which was undertaken at the age of 3–6 months. This was followed by primary palatoplasty at approximately 12 months of age. Gingivoperiosteoplasty was not performed. After primary surgery, a residual unrepaired alveolar cleft was present, which was to be reconstructed by subsequent SABG in the mixed dentition. Pre-SABG expansion was performed for patients who presented with maxillary constriction, posterior crossbite, or asymmetrical arch form and/or for those who required better access for graft placement. SABG was performed either prior to or soon after the emergence of the CS permanent maxillary canine through the alveolar bone or mucosa, with the former being the preferred timing when possible.

To ensure sample homogeneity and to reduce surgical variation, patient inclusion criteria included only patients with cUCLP who had all cleft-related surgeries and SABG performed by the same experienced surgeon (Dr Fisher), had received no other surgical procedures during the observation period, and had suitable radiographic records to visualize root development of the maxillary central incisors and canines on the CS and NCS. Patients with craniofacial syndromes, missing radiographic records, or poor visualization of the teeth on radiographs to allow determination of their root development status and those with Simonart's band or soft tissue bridging were excluded.

Panoramic or periapical radiographs were collected and analyzed at three time points: time point 1 (T1), pre-SABG (mean age, 7.55 years); T2, at the time of SABG (immediately before or up to 3 months before SABG; mean age, 10.13 years); and T3, post-SABG (at least 2 years after SABG; mean age, 14.19 years). The longitudinal root development of the CS and NCS permanent maxillary canines and central incisors was analyzed from the radiographs using the rating system of El Deeb et al.[6] (Table 2; Figure 1), which compares relative root lengths and crown heights. All root development scoring was performed by the same investigator (Dr Vandersluis-Solomon). As root development can be affected by interpretation, intrarater reliability was assessed through the random selection and analysis of 13 radiographs at each time point 1 month after the initial assessment.

Statistical analysis included descriptive statistics, paired *t*-tests, and analyses of proportions. The Wilcoxon signed-rank test was used to determine if the

Table 5. Distribution of Root Development Scores of NCS and CS Maxillary Central Incisors and Canines Observed at Each Time Point[a]

		Root Development Score			
		Central Incisor		Canine	
Time Point	Score	NCS, %	CS, %	NCS, %	CS, %
T1 (7.63 ± 0.91 y)	0	0.00	0.00	2.44	6.98
	1	2.38	2.38	31.71	37.21
	2	33.33	33.33	56.10	51.16
	3	11.90	9.52	4.88	2.33
	4	40.48	45.24	4.88	2.33
	5	7.14	9.52	0.00	0.00
	6	4.76	0.00	0.00	0.00
T2 (10.11 ± 0.77 y)	1	0.00	0.00	0.00	0.00
	2	0.00	2.50	11.76	24.39
	3	0.00	0.00	29.41	34.15
	4	10.53	17.50	50.00	39.02
	5	34.21	45.00	5.88	0.00
	6	55.26	35.00	2.94	2.44
T3 (14.14 ± 1.62 y)	1	0.00	0.00	0.00	0.00
	2	0.00	0.00	0.00	0.00
	3	0.00	0.00	0.00	0.00
	4	0.00	0.00	4.89	18.60
	5	0.00	7.14	7.32	6.98
	6	100.00	92.86	87.80	74.42

[a] NCS indicates noncleft side; CS, cleft side; T1, time point 1; T2, time point 2; and T3, time point 3.

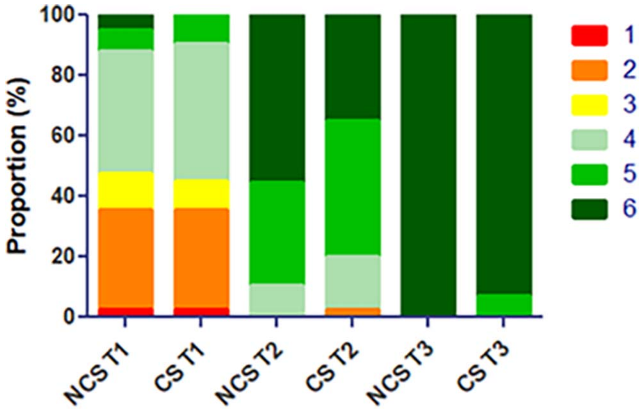

Figure 2. Root development scores of the maxillary central incisor observed at different time points. NCS indicates noncleft side; CS, cleft side; T1, time point 1; T2, time point 2; and T3, time point 3.

differences in the root development scores were significant at the respective time points. Regression modeling of the CS and NCS permanent maxillary central incisor and canine root development scores as a function of age was also undertaken.

RESULTS

After applying all inclusion and exclusion criteria, a total of 44 subjects with cUCLP who had all cleft-related and SABG surgeries performed by the same surgeon were selected. This sample size was similar to those of other published studies that looked at the effects of SABG on root development in cUCLP (Table 1). Sample characteristics are shown in Table 3. The intrarater reliability for root development scoring was excellent (k = 0.85; $P < .001$). Root development of the CS and NCS permanent maxillary central incisors and canines was not adversely affected by SABG as all teeth reached at least stage 4 of root development, and most reached stage 5 or 6, at T3.

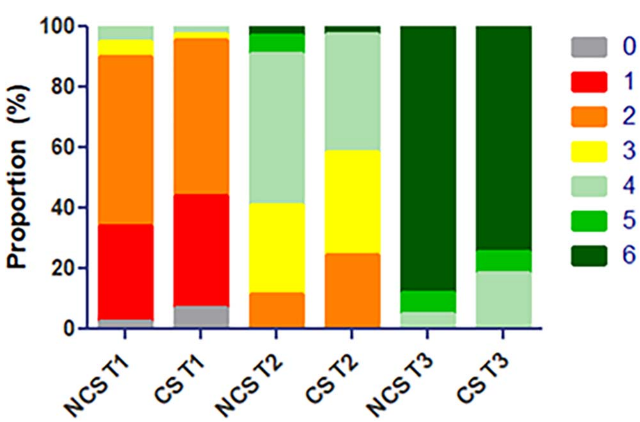

Figure 3. Root development scores of the maxillary canine observed at different time points. NCS indicates noncleft side; CS, cleft side; T1, time point 1; T2, time point 2; and T3, time point 3.

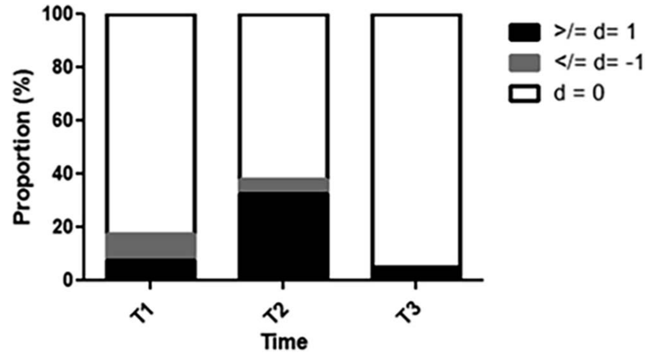

Figure 4. Proportion of maxillary central incisors at each time point with a difference in the root development score of at least 1 between the CS and NCS or no difference. The root development score difference was calculated as NCS − CS for all time points, where >/= d = 1 indicates that the NCS score is higher by at least 1, </= d = −1 indicates that the CS score is higher by at least 1, and d = 0 indicates no difference between the NCS and CS scores. T1 indicates time point 1; T2, time point 2; and T3, time point 3.

For both the maxillary central incisor and the canine, the root development of the CS tooth was delayed relative to the NCS tooth, with the greatest difference being seen at T2 (Tables 4 and 5; Figures 2 and 3). A larger proportion of teeth on the CS trailed those on the NCS by at least 1 point at T2 than at T1 or T3, with the smallest proportion being seen at T3 (Figures 4 and 5). Regression modeling of the CS and NCS permanent maxillary central incisor and canine root development scores as a function of age[20] revealed that the average root development score differences for the canine and the central incisor were the greatest at T2 but diminished at T3 (Figures 6 and 7). An examination of the slopes of the graphs revealed that the greatest acceleration of root development for the maxillary central incisors generally occurred between T1 and T2. Examination of the slopes of the

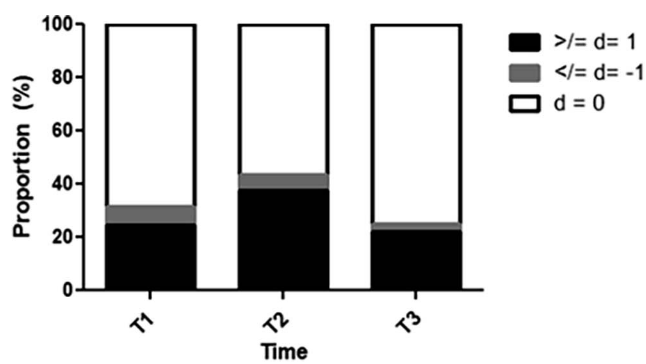

Figure 5. Proportion of maxillary canines at each time point with a difference in the root development score of at least 1 between the CS and NCS or no difference. The root development score difference was calculated as NCS − CS for all time points, where >/= d = 1 indicates that the NCS score is higher by at least 1, </= d = −1 indicates that the CS score is higher by at least 1, and d = 0 indicates no difference between the NCS and CS scores. T1 indicates time point 1; T2, time point 2; and T3, time point 3.

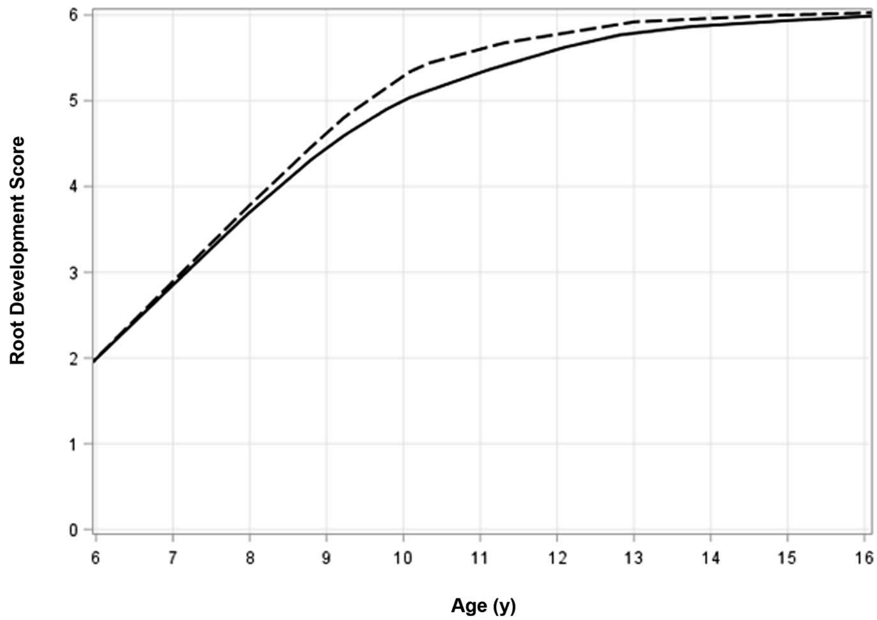

Figure 6. Regression modeling of the cleft and noncleft permanent maxillary central incisor root development scores as a function of age. A local regression line was fitted for each group to depict the average root development over time (shown for ages 6 years to 16 years). Solid line, CS; Dashed line, NCS.

graphs of the maxillary central incisors and canines on the CS and NCS from T1 to T2 and from T2 to T3 revealed a relative CS acceleration from T2 to T3, indicating a catch-up of root development of cleft-adjacent teeth after SABG (Figures 6 and 7).

DISCUSSION

Cleft-adjacent teeth have more frequent agenesis, tooth shape and size variations, eruption delays,[1,3,7,8,24–27] poor periodontal support,[28–31] and caries risk.[31,32] With a high frequency of agenesis of maxillary lateral incisors on the CS, the maxillary central incisors and canines are frequently cleft-adjacent teeth, and clinicians need to observe differences in their shape, size, and development compared with their contralateral analogs, especially when planning and executing the reconstruction of the alveolar cleft by SABG, orthodontic correction of the dentition in this area, and/or restorative treatment for esthetics and long-term health and function.

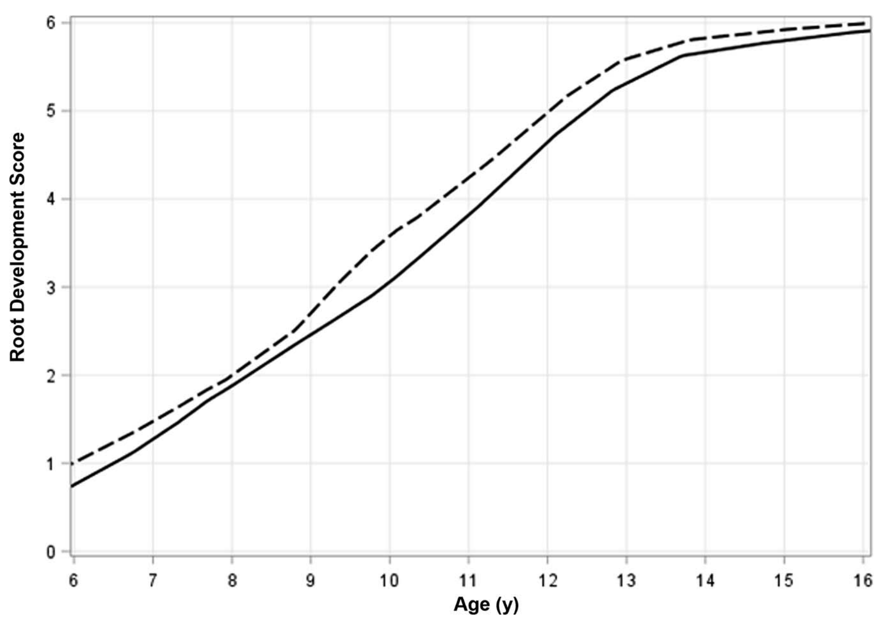

Figure 7. Regression modeling of the cleft and noncleft permanent maxillary canine root development scores as a function of age. A local regression line was fitted for each group to depict the average root development over time (shown for ages 6 years to 16 years). Solid line, CS; Dashed line, NCS.

The literature regarding the root development of cleft-adjacent teeth following SABG in UCLP is equivocal. Some studies reported a delay in maxillary canine root development on the CS that showed an acceleration after SABG,[10,18,20–22] consistent with the findings of this investigation. Others found no significant differences.[6,19] Regarding the maxillary central incisors, some reports described a delay in root development at the time of SABG,[20] while others did not.[10] In this investigation, delayed root development was seen more frequently on the CS for both the maxillary central incisors and maxillary canines. Results of *t*-tests and Wilcoxon signed-rank tests showed significantly lower root development scores ($P < .05$) at T2 for the CS central incisors and at all three time points for the CS canines (Table 3; Figures 2 through 5). However, a catch-up or acceleration was evident, especially in the CS canines, roughly around and after the age of 10 years, corresponding to T2 to T3 (Figures 6 and 7), indicating that there was likely a favorable effect of SABG on the root development of the CS maxillary canine with the provision of mesenchymal matrix and bone volume at the cleft site.

While this was a retrospective longitudinal study using radiographs that had been acquired to support clinical decision-making and follow-up, all cleft surgeries and SABG were conducted by the same experienced plastic surgeon using the same standardized technique. This improved sample homogeneity regarding the surgical aspects of treatment that all patients in the sample received. The findings of this study clarified that the development of the cleft-adjacent teeth was not adversely affected by SABG. The provision of the alveolar bone graft soon after T2 allowed cleft-adjacent teeth that were slower in root development to catch up with their contralateral analogs. This supports the recommendation of providing SABG to patients with cUCLP from the standpoint of promoting the root development of the cleft-adjacent teeth. Ethical constraints precluded the possibility of having a control group that would not have had SABG, and there was no equivalent sample that had longitudinal radiographs at similar ages as the included study patients but had not been provided with bone grafts. This limits the possibility of concluding that the catch-up in root development seen in the CS teeth after SABG would not have happened or would have happened at a different rate without SABG.

Three-dimensional (3D) and cone-beam computed tomography (CBCT) images at various time points were not available due to radiation exposure and costs. Future studies could incorporate 3D imaging when available to better understand any aspects of the grafting process that may enhance or affect root development. While statistically significant and frequent, the mean magnitude of the differences in the root development scores was modest. This implies that clinicians should observe each patient's root development on the CS and NCS carefully for alveolar bone grafting.

CONCLUSIONS

- Root development of cleft-adjacent central incisors and canines is slow in comparison to their noncleft analogs.
- Root development of these teeth accelerates following SABG.

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