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Differences in the panoramic appearance of cleft alveolus patients with or without a cleft palate

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ABSTRACT

Purpose: The purpose of this study was to clarify the panoramic image differences of cleft alveolus patients with or without a cleft palate, with emphases on the visibility of the line formed by the junction between the nasal septum and nasal floor (the upper line) and the appearances of the maxillary lateral incisor.

Materials and Methods: Panoramic radiographs of 238 patients with cleft alveolus were analyzed for the visibility of the upper line, including clear, obscure or invisible, and the appearances of the maxillary lateral incisor, regarding congenital absence, incomplete growth, delayed eruption and medial inclination. Differences in the distribution ratio of these visibility and appearances were verified between the patients with and without a cleft palate using the chi-square test.

Results: There was a significant difference in the visibility distribution of the upper line between the patients with and without a cleft palate ($p<0.05$). In most of the patients with a cleft palate, the upper line was not observed. In the unilateral cleft alveolus patients, the medial inclination of the maxillary lateral incisor was more frequently observed in patients with a cleft palate than in patients without a cleft palate.

Conclusion: Two differences were identified in panoramic appearances. The first was the disappearance (invisible appearance) of the upper line in patients with a cleft palate, and the second was a change in the medial inclination on the affected side maxillary lateral incisor in unilateral cleft alveolus patients with a cleft palate. ***(Imaging Sci Dent 2024; 54: 25-31)***

KEY WORDS: Jaw Abnormalities; Cleft Palate; Radiography, Panoramic

Introduction

A cleft lip is one of the most frequently observed congenital anomalies in the maxillofacial region, and the majority of patients have a cleft alveolus and/or a cleft palate.[1] Bone transplantation is performed as an effective treatment for alveolar defects of patients that are 8-10 years old.[2,3] Patients are followed up on a regular basis by physical and imaging examinations to ensure sufficient maxilla growth prior to surgery. Panoramic radiography plays an essential role in such repeated examinations because of its low cost and low radiation exposure.[4]

However, it is difficult for oral and maxillofacial radiologists (that routinely interpret radiographs and create reports in hospital radiology departments) to physically examine all patients. Therefore, the radiologists are frequently required to estimate the patients' physical characteristics using only radiographic appearances. In this regard, panoramic radiography is an effective method because it can be used to visualize the entire jaw and the surrounding structures on one image. Although diagnosis of the presence of a cleft alveolus is usually not an issue for experienced radiologists, it is sometimes difficult for inexperienced observers.[5,6] Correct differentiation of a cleft alveolus without a cleft palate from those with a cleft palate may not be easy, even for experienced radiologists.[5,7] There have been numerous studies

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clarifying the radiographic features of cleft alveolus patients, including the appearances of tooth and alveolar abnormalities on panoramic radiographs, and some differences from healthy subjects have been reported.[8-11] Compared with other permanent teeth, the maxillary lateral incisor most frequently shows anomalies such as it is missing, has incomplete or delayed development in the crown and/or root, medial inclination, and delayed eruption. Although these anomalies are observed more frequently in the cleft side than in the contralateral side in patients with unilateral cleft alveolus, the differences between the patients with and without a cleft palate are not completely clarified.[8-11]

Although the hard palate and nasal floor of cleft palate patients are naturally affected, their appearances on panoramic radiographs have not been investigated using the principle of panoramic image formation. When a healthy subject is set in an ordinary positioning in which the Frankfurt horizontal (FH) plane is parallel to the floor, two horizontal lines (the upper and lower lines) are visible in the palatal regions on the panoramic radiographs.[12,13] Damante et al.[14] and Azevedo and Damante[15] clarified the composition of these appearances in experimental studies. The upper line is mainly formed by the junction between the nasal septum and nasal floor and the posterior portion of the hard palate and nasal floor, while the lower line is formed by the nasal floor, especially its lateral and anterior limits. In patients with a cleft palate, the junctions of the nasal septum and nasal floor are usually affected and they disappear bilaterally or unilaterally. Therefore, the appearance of the upper line changes. In patients with a cleft palate, especially in the case of a cleft palate connecting to a cleft alveolus, the lines are more severely affected.

The aim of this study was to clarify the differences in panoramic image findings of cleft alveolus patients with or without a cleft palate, with emphasis on the visibility of the upper line formed by the junction between the nasal septum and nasal floor, and the appearances of the maxillary lateral incisors.

Materials and Methods

The study was approved by the ethics committee of our university (No. 649) and performed according to the principles of the Declaration of Helsinki. Because of the noninvasive and retrospective natures of this observational study using only existing anonymized image data, the ethics committee waived the requirement for informed consent from the participants. All patients were given the opportunity to refuse to participate in the study by declaring to opt-out.

Subjects

Panoramic radiographs of 238 patients (105 females and 133 males) between August 2004 and July 2020 with unilateral (200 patients) or bilateral (38 patients) cleft alveolus were selected from our hospital image database. Among several panoramic examinations repeated for a patient, one immediately before bone graft surgery was selected. The mean age was 9.03 years. In the unilateral cleft alveolus group, 100 patients had a cleft palate and the remaining 100 patients did not have a cleft palate. The former was assigned as the "with cleft palate" group, and the latter was the "without cleft palate" group. In the bilateral group, 18 patients were associated without a cleft palate ("without cleft palate" group). The presence or absence of a cleft alveolus and cleft palate was verified with the medical records and computed tomographic (CT) appearances.

All panoramic images were obtained digitally by a Vera viewepocs (J Morita Corp, Kyoto, Japan), which X-ray tube was changed twice, with a tube voltage of 75 kVp, tube current of 8 mA, and exposure time of 16.2 seconds.

Image analysis

The panoramic images were downloaded from the image database as a digital imaging and communication in medicine (DICOM) format and prepared for interpreting the images. We manually excluded the images in which the alveolar bone, inferior margin of the piriform aperture and incisor of the maxillary anterior teeth could not be clearly observed. Two oral and maxillofacial radiologists with 8 (CK) and 38 (EA) years' experience interpreted the panoramic image appearances on a high resolution 21.3-inch monitor (RadiForce RX240 monitor, EIZO Corporation, Hakusan, Japan) with 1200 × 1200 pixel resolution and 8-bit RGB support. The radiologists separately evaluated two aspects of the image features without any clinical information, except for whether the patients had a unilateral or bilateral cleft alveolus.

The first aspect evaluated was the upper line formed by the nasal floor and nasal septum (Fig. 1). When the upper line was clearly observed on both the right and left side, it was classified as "Clear" (Fig. 1A). When the line was partially or unclearly observed, it was classified as "Obscure" (Figs. 1B and C). If the upper line was not observed, it was assigned as "Invisible" (Figs. 1D and E).

The second aspect was the appearances of the lateral incisor related to the cleft alveolus area (Fig. 2). For the unilateral cleft alveolus group, the radiologists initially evaluated whether the incisor was present or absent (Fig. 2A). Subsequently, for the present incisors, visible or invisible incomplete growth in the crown and/or root, including micro-

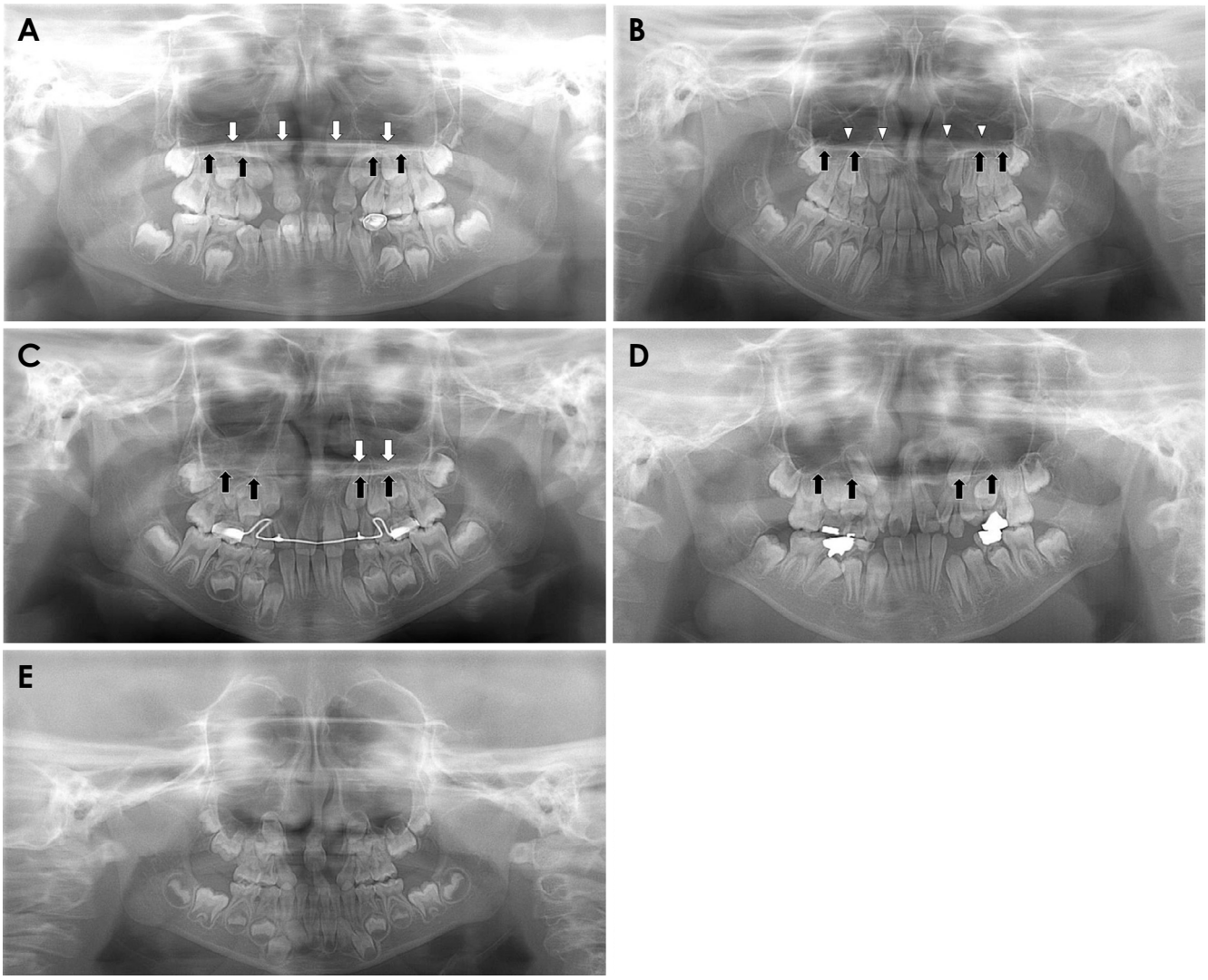

Fig. 1. The upper lines in "Clear" (A), "Obscure" (B and C), and "Invisible" (D and E) appearances. A. The upper line is clearly visible at the nasal and maxillary sinus floors levels (white arrows), as well as the lower line (black arrows). B. The upper line is observed to be unclear on both the right and left sides (white arrowheads) compared to the lower line (black arrows), however the line is slightly observable. C. The left side upper line (white arrows) is observed as well as the lower line (black arrows); however, it is invisible on the right side. D. The lower line is visible (black arrows), while the upper line is not observed on either side. E. The upper and lower line are not observed on either side.

dontia (Fig. 2B), delayed eruption (Fig. 2C), and medial inclination (Fig. 2D), were evaluated. For the bilateral cleft alveolus group, the presence or absence of the incisor was assigned as three categories, present bilaterally, present unilaterally, and absent bilaterally. For existing incisors, incomplete growth in the crown and/or root, including microdontia, delayed eruption, and medial inclination, were assessed by classifying as the following five categories, 1) invisible bilaterally with both side incisors existing, 2) visible unilaterally with both side incisors existing, 3) visible bilaterally with both side incisors existing, 4) visible unilaterally with one side incisor existing, and 5) invisible with one side incisor existing.

When the evaluations by the radiologists were different, final decisions were reached by consensus after discussion. Before the final decisions of above appearances, the inter-observer agreements were evaluated between two observers based on Cohen's kappa statistics modified for multi-categorized data.[16] The agreements were classified as poor when $\kappa < 0.2$, fair when $0.2 \leq \kappa < 0.4$, moderate when $0.4 \leq \kappa < 0.6$, good when $0.6 \leq \kappa < 0.8$, and very good when $0.8 \leq \kappa$.[17]

Differences in the distribution ratio of the panoramic appearances were verified between the cleft alveolus patients with or without a cleft palate using the chi-square test with a significance level less than 0.05.

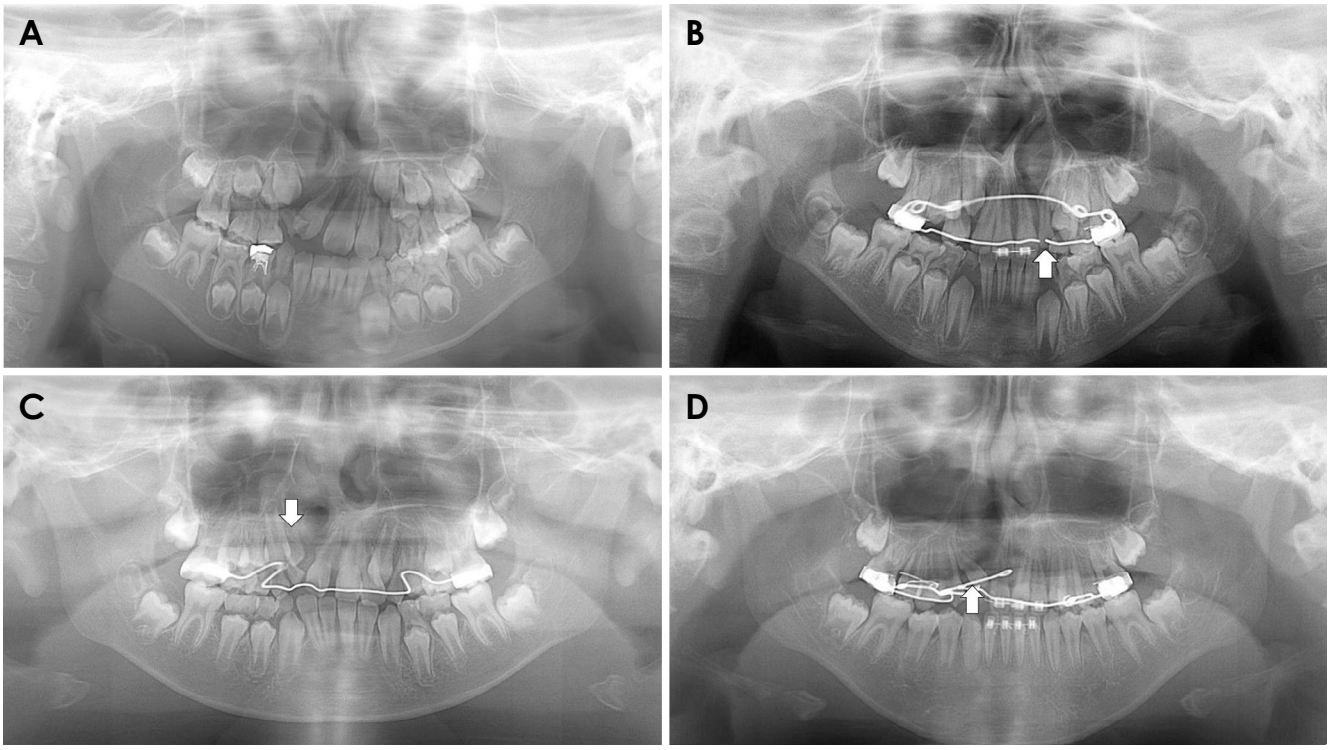

Fig. 2. Appearances of the maxillary lateral incisor. A. The right lateral incisor cannot be observed and assigned as "absent". B. The left lateral incisor (arrow) is assigned as "incomplete growth (microdontia)" because it is obvious when it is compared with the right lateral incisor. C. The right lateral incisor (arrow) is not erupted and assigned as "delayed eruption" while the left lateral incisor is completely erupted. D. The right lateral incisor (arrow) is assigned as "medial inclination".

Table 1. Visibility of the line formed by the nasal floor and nasal septum

Cleft alveolus status	Group	Appearance			p*	Inter-observer agreements
		Clear	Obscure	Invisible		
Unilateral cleft alveolus	With cleft palate	0	17	83	<0.05	0.95
	Without cleft palate	83	10	7		
Bilateral cleft alveolus	With cleft palate	2	5	13	<0.05	0.54
	Without cleft palate	16	0	2		

*: difference in the distribution ratios of visibility between groups

Results

The visibilities of the upper line is summarized in Table 1. The observer agreement (kappa values) of unilateral and bilateral group was 0.95 and 0.54, respectively. The unilateral group was classified as very good, while the bilateral group was classified as moderate. There was a significant difference in the distributions of visibilities between the groups with and without a cleft palate for both unilateral and bilateral cleft alveolus patients ($p<0.05$). In 83% of the unilateral cleft alveolus patients without a cleft palate, the upper line was clearly visible, while it was invisible or obscured in all patients with a cleft palate. In the bilateral group, 16 of 18 (89%) patients without cleft palate were classified as Clear, while 18 of 20 (90%) patients showed the Obscure or Invisible appearance.

The appearances of the lateral incisor are summarized in Tables 2 and 3. The agreement of two observers (kappa value) regarding the present or absent of incomplete growth was 0.60 (good consistency) in the unilateral cleft alveolus group, while all others were classified as very good. The appearances of the lateral incisors were not different for the

Table 2. Appearances of the lateral incisor in unilateral cleft alveolus patients

Appearance		With cleft palate	Without cleft palate	p*	Inter-observer agreements
Present or absent	Present	58	68	0.143	0.88
	Absent	42	32		
Incomplete growth	Visible	44	57	0.264	0.60
	Invisible	14	11		
Delayed eruption	Visible	44	49	0.628	0.85
	Invisible	14	19		
Medial inclination	Visible	26	13	<0.05	0.85
	Invisible	32	55		

*: difference in the distribution ratios of appearance between groups

Table 3. Appearances of the lateral incisor in bilateral cleft alveolus patients

Appearance	With cleft palate	Without cleft palate	p*	Inter-observer agreements
Present or absent				
Present bilaterally	12	11		
Present unilaterally	4	1	0.342	0.85
Absent bilaterally	4	6		
Incomplete growth				
Invisible bilaterally with both side incisors existing	8	4		
Visible unilaterally with both side incisors existing	1	0		
Visible bilaterally with both side incisors existing	3	7	0.242	0.85
Visible with one side incisor existing	3	1		
Invisible with one side incisor existing	1	0		
Delayed eruption				
Invisible bilaterally with both side incisors existing	2	0		
Visible unilaterally with both side incisors existing	1	2		
Visible bilaterally with both side incisors existing	9	9	0.141	0.82
Visible with one side incisor existing	4	0		
Invisible with one side incisor existing	0	1		
Medial inclination				
Invisible bilaterally with both side incisors existing	4	7		
Visible unilaterally with both side incisors existing	3	2		
Visible bilaterally with both side incisors existing	5	2	0.305	0.94
Visible with one side incisor existing	1	1		
Invisible with one side incisor existing	3	0		

*: difference in the distribution ratios of appearance between groups

groups with or without a cleft palate, except for the medial inclination in unilateral cleft alveolus patients ($p<0.05$) (Tables 2 and 3). In 26 of 58 (45%) unilateral cleft alveolus patients with cleft palate, the medial inclination of the lateral incisor was observed, while only 13 of 69 (19%) patients without cleft palate showed the medial inclination.

Discussion

Although CT and cone-beam CT are effective tools for investigating the cleft alveolus patients, panoramic radiography still has a role because of its low cost and low radiation exposure.[18]

The panoramic appearances constructed by the bony structures surrounding the nasal cavity and hard palate have been reported by several investigators. In the ordinary position with the FH plane parallel to the floor, two horizontal lines are usually observed at approximately the level of the nasal floor and maxillary sinus floor between the bilateral posterior wall lines of the maxillary sinus. A previous publication indicated that "The hard palate appears as two radiopaque lines. The lower line represents the junction between the hard palate and lateral nasal wall on the receptor side of the patient. The upper line represents the junction between the nasal wall and hard palate on the tube side or ghost image of the opposite side".[12] In 1987, Reijnen and Sanderink[13] concluded that the lower line was created by the junction of the maxillary sinus wall and the hard palate, and it was always clear, while the upper line produced by the junction between the nasal septum and nasal floor was always blurred when it appeared. The two lines sometimes superimposed when a patient was set in the chin-up position. In 1998 and 2001, Damante et al.[14] and Azevedo and Damante[15] experimented with dried human skulls in which bony defects similar to a cleft palate were created artificially. Their results were similar to those of Reijnen and Sanderink.[13] The upper line did not appear in the panoramic images taken with the dried maxilla with a cleft palate -like bony defect. Therefore, the appearances of the upper line change in patients with a cleft palate. However, such changes have not been investigated in relation to the existence of a cleft palate. In this regard, the hypothesis in this study was that the upper line could be clearly visible in cleft alveolus patients without a cleft palate, while it would not be visible in patients with a cleft palate. Although the hypothesis could be verified with these results, there were numerous cases of the "Obscure" appearance, which may be attributed to the status of the junctions between the nasal floor and nasal septum. This contact status might vary individually. In a patient with cleft palate, the nasal septum may not be in contact with the nasal floor. In another patient, the nasal septum may be attached to either the right or left side of the separated nasal floor. Moreover, both structures may be partially contacted in the antero-posterior direction. The observer agreement for the appearance of the upper line in the bilateral cleft alveolus group was lower (0.54) than the unilateral cleft alveolus group. This might be related to the fact that the relationship between the nasal septum and nasal floor was more complex and that there were fewer cases than unilateral cleft alveolus group. Therefore, this results bilateral cases may require a further analysis. The constitution of the upper line on panoramic image should be analyzed in detail with reconstructed CT features in a future study. The "Invisible" appearance of patients without a cleft palate might be attributed to their chin-up position, resulting in the appearance of one superimposed line.

There are a few appearances that differentiate the patients with a cleft palate from those without a cleft palate. The supernumerary tooth is more frequently observed in cleft alveolus patients without a cleft palate than in those with a cleft palate.[11] The incidence of cross bite is greater in cleft alveolus patients with a cleft palate than those without a cleft palate.[11] However, regarding the maxillary lateral incisors, effective appearances have not been reported. In this study, the medial inclination increased for unilateral cleft alveolus patients with a cleft palate. The enlargement of the bony defect, which was created by the connection of the cleft alveolus and a cleft palate, may be related to the change in the number of medial inclination incisors in cleft alveolus patients with a cleft palate.

In this study, the unilateral or bilateral cleft alveolus patients were separately evaluated because the appearances and number of bilateral cleft alveolus patients were considerably different from those of unilateral cleft alveolus patients. Moreover, we can easily differentiate between these two patients based on panoramic appearances. For bilateral cleft alveolus patients, the results should be confirmed with more patients, especially for the appearance of the maxillary lateral incisor.

We previously investigated the use of a deep learning model for differentiating the cleft alveolus cases with a cleft palate from those without a cleft palate using panoramic radiographs.[7] Generally, the performance of the DL system for image classification is improved when smaller areas are used.[19,20] We compared the classification performances from small areas of the maxillary incisor regions to those from the whole panoramic area.[7] Contrary to our expectations, a high performance was verified both for the images. Therefore, we expected a definitive appearance that occupied a relatively wide area on the panoramic images and could differentiate the subjects with a cleft palate from those without; however, it was not sufficiently identified by human observers.

In conclusion, we clarified two differences between cleft alveolus patients with a cleft palate and those without. The first was the disappearance (invisible appearance) of the upper line formed by the nasal floor and nasal septum in patients with cleft palate, and the second was the occurrence ratio change of medial inclinations in the affected side maxillary lateral incisor in unilateral cleft alveolus patients with a cleft palate.

Conflicts of Interest: None

Acknowledgments

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