Treatment of identical twins with congenitally missing maxillary lateral incisors – a long-term follow-up

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Congenitally missing upper lateral incisors can significantly alter the aesthetic appearance of a smile. The orthodontic challenge is to achieve acceptable facial aesthetics by restoring the dentition either by replacing the missing teeth or by substituting the canines for the lateral incisors. The present case report discusses the orthodontic treatment of two identical twin sisters with missing upper lateral incisors, in whom the spaces were reopened to accommodate prosthetic replacement. The rationale for this treatment alternative is discussed.

Introduction
Hypodontia is defined as the developmental absence of one or more teeth.1 In many populations, it has been reported that, after the third molars, the most frequently, congenitally missing teeth are the maxillary lateral incisors.2 In Caucasian populations, lateral incisor agenesis has a prevalence of 1–2%, the incidence being higher when other family members are affected.3 In addition, it has been established that the condition is 1.5 times more common in females than males.4

In essence, there are two orthodontic treatment options to manage congenitally missing lateral incisors. Option one is to close the created space by substituting the canines for the lateral incisors, combined with reshaping teeth for improved aesthetics.5-9 The alternative treatment involves reopening space for the prosthetic replacement of the lateral incisors.10

Selecting the appropriate treatment approach is guided by the patient’s existing malocclusion, growth pattern, and any potential adverse impact on the patient’s anterior aesthetics.

Diagnosis and etiology

History
M. and L. were monozygotic twin sisters. They were 12 years old at the beginning of treatment, and were in the late mixed dentition. Clinical and radiographic examinations revealed congenitally missing upper lateral incisors (Figures 1 – 3). The permanent maxillary canines erupted mesially to the retained deciduous canines. The mother also had missing upper lateral incisors, reinforcing the familial pattern of the condition.11 The girls were otherwise healthy.

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Clinical examination – extra-oral and intraoral

The sisters had symmetrical, slightly dolichocephalic faces (Figure 1). Their facial profiles were convex, highlighted by mandibular retrusion and an increased naso-labial angle. In the smiling photographs, excessive gingival display and wide buccal corridors may be noted.

The dental pattern in both sisters was a Class I malocclusion with excessive overbite and overjet. The deciduous upper canines were retained, while the permanent canines had adopted the position of the congenitally missing lateral incisors. M. also had the right second mandibular primary molar still present, while L. had a retained left second mandibular primary molar. Mild crowding existed in both arches, as well as an accentuated lower curve of Spee. However, the upper arch had a reverse curve due to the overeruption of the central incisors. The periodontal condition was within normal limits and generally healthy.
The maxillary lateral incisors were congenitally missing in both girls (Figures 3 and 4). The deciduous canines had partially resorbed roots and the remaining primary molars were in the process of exfoliating. The condyles were symmetrical and appeared normal. No other pathology was noted.

Cephalometric radiographs and tracings
The cephalometric radiographs (Figure 5) and analyses (Figure 6; Table I) revealed that the twins had a moderate Class II skeletal pattern due to mandibular retrusion with a hyperdivergent component. The maxillae (relative to the cranial base) were in the normal range (SNA – M.: 82°; L.: 80°), while their mandibles were retrognathic (SNB – M.: 76°; L.: 75°). The difference in the skeletal bases (ANB) showed a similar skeletal pattern (ANB – M.: 5.5°; L.: 5°). The inclination of the upper incisors relative to the Frankfort Horizontal plane was approximately 10° less than normal for both sisters, indicating retroclined upper anterior teeth. The lower incisors relative to the mandibular plane were slightly retroclined for M. at 85°, but slightly proclined for L. at 94°. The twins had elevated mandibular plane angles (FMA – M.: 33.7°; L.: 33.1°) and their lower anterior face heights indicated a hyperdivergent growth pattern (LAFH – M.: 65.1mm; L.: 63.1mm).
Table I. Cephalometric values for M. and L. at T1 (12Y 1M) and T2 (14Y 10M).

| Metric                      | T1 – 12Y 1M | Norm | T2 – 14Y 10M | Norm |
|-----------------------------|-------------|------|--------------|------|
| SNA                         | 81.7°       | 80.0°| 78.2°        | 77.8°|
| SNB                         | 76.2°       | 74.9°| 74.4°        | 73.9°|
| ANB                         | 5.5°        | 5.1° | 2            | 3.8° |
| 1/ (FH)                     | 101.9°      | 98.6°| 112.6°       | 110.5°|
| IMPA                        | 85.0°       | 94.2°| 97.3°        | 102.8°|
| L / T                       | 139.4°      | 134.2°| 132.1        | 113.8°|
| VITSM                       | -0.8mm      | -3.2mm| -1.4         | -2.2mm|
| FMA                         | 33.7°       | 33.1°| 26.4         | 36.4° |
| LL – E plane                | -1.0mm      | -1.6mm| 0.5          | -0.7mm|
| LAFH                        | 65.1mm      | 63.1mm| 53.9         | 71.0mm|

Figure 5. Pretreatment lateral cephalometric radiographs.

Figure 6. Pretreatment lateral cephalometric tracings.

**Treatment objectives**

1. To improve facial and dental aesthetics.
2. To open spaces for the prosthetic replacement of the maxillary lateral incisors.
3. To obtain a Class I dental relationship.
4. To reduce overbite and overjet.

**Treatment alternatives**

Two alternatives exist for the treatment of tooth agenesis. In the case of missing maxillary lateral incisors the first option is space closure by protraction of the canines and buccal segments, resulting in a therapeutic Class II molar relationship. If the...
Due to the twins’ more obtuse naso-labial angle, it was considered desirable to advance, or at least maintain, the position of the maxillary incisors, to avoid retraction of the upper lip as a result of space closure. Therefore, the agreed treatment option was to reopen the spaces for prosthetic replacement of the upper lateral incisors. This would permit the maintenance of a Class I relationship, since no extractions were deemed necessary in the mandibular arch.

Obtaining the required spaces for the upper lateral incisors was achievable with a combined approach involving the extraction of the deciduous canines, expansion of the arch, molar distalisation and proclination of the central incisors.

**Treatment progress**

The remaining deciduous teeth were extracted. In order to re-establish appropriate arch width, a banded rapid palatal expansion appliance (Haas Type) was placed, resulting in approximately five millimetres of widening in each patient’s maxilla (Figure 7). Fixed edgewise brackets (0.022” × 0.028” slot) were bonded to the upper teeth, and open coil springs were
placed to move the upper canines distally into their proper positions and thereby re-open spaces for the upper lateral incisors (Figure 8). Retraction springs were also used from the canines to the maxillary first molars. Conventional fixed appliance treatment for alignment was performed on the mandibular arch.

To compensate for the backward rotation of the mandible due to maxillary expansion, some distalisation of the upper posterior teeth was necessary to re-establish Class I interdigitation. A removable appliance with finger springs against the molars and a headgear were used to distalise the molars, followed by retraction of the premolars and canines using a second removable appliance and Class II elastics. Since considerable anterior spaces were developed, prosthetic teeth were added to the removable appliance to replace the lateral incisors.

Active treatment time spanned 30 months, and at age fourteen a fixed lingual retention wire was bonded between the maxillary central incisors, a Hawley appliance with prosthetic lateral incisors was placed on the maxillary arch (Figure 9) and a removable retainer in the lower arch.

The twins found it difficult to tolerate the removable upper retainers while eating, and so these were replaced by fixed retention with pontics on a lingual arch anchored to bands on the upper first molars (Figure 10). The twins found these were preferable and they were maintained very well.

Treatment results

The post-treatment intraoral photographs (Figure 9) and models (Figure 11) show that a super Class I molar relationship and good overbite and overjet were obtained for both sisters. The occlusal views indicate that a more rounded arch form was developed. The post-treatment panoramic radiographs (Figure 12) show little or no root resorption of the maxillary and mandibular anterior teeth with good root parallelism of the adjacent teeth. A space of six millimetres was maintained for each lateral incisor.

Cephalometric results

The maxillary bases were retracted in both cases (SNA – M.:82° → 78°; L.: 80° → 78°). The mandibular bases were relatively stable (SNB – M.:76° → 74°; L.:75°...
The maxillary incisors were proclined to more normal values in both patients (1/FH – M.: 102° → 113°; L.: 99° → 111°). However, there was a slight difference in the degree of proclination of the lower incisors to the mandibular plane (IMPA – M.: 85° → 97°; L.: 94° → 103°). The greatest differences noted were an increase in lower anterior face height (LAFH – M.: 65 → 71 mm; L.: 63 → 68 mm).

The cephalometric superpositions (Figure 15) show that M. had a slightly greater rotation of the mandible in a downward and backward direction compared with L.

The extra-oral photographs (Figure 10) show that the frontal views have remained unchanged and that the profile views have remained slightly retrognathic. There was a dramatic improvement in their smiling views with respect to a reduction in both gingival display and buccal corridors.
At the age of 21 both sisters were ready for definitive replacement of the lateral incisors. They chose for the placement of Maryland bridges, which yielded a highly aesthetic result for both (Figure 16). The alignment of the lower anterior teeth in Twin L. demonstrated some relapse since her lower lingual bonded retainer was no longer present.

At a review visit (15 years after the completion of treatment), the twins indicated that they had recently had the Maryland bridges replaced by implants. It is likely that their dentist had suggested this treatment alternative following repeated recementation of the detached Maryland bridges. While the implant site for the left lateral incisor in Twin L. was slightly narrower, there was still adequate space to accommodate the implant. An examination of the sisters (at age 29) showed some differences with respect to the aesthetics of the final result (Figure 17). There was a marked apical gingival migration around the implants in Twin L., as well as a marked rotational relapse of the lower incisors and some change in the upper central incisors. Retention compliance was generally an issue with
Twin L., who did not wear her appliances as diligently as Twin M., particularly following the removal of her lower anterior bonded retainer.

Discussion

In managing bilateral agenesis of the maxillary lateral incisors, proper treatment planning is paramount to providing an acceptable aesthetic result.

According to Zachrisson et al.,⁹ conventional space closure for missing lateral incisors is a viable and safe procedure that provides satisfactory aesthetic and functional long-term results. A recent case report by de Almeida et al.¹⁴ supports this view, referring to the maintenance of stability in a 14-year follow-up of a patient treated by space closure. It seems, however, that equally successful long-term stability may be achieved in cases in which the alternative option of space-opening was selected. Kokich et al.¹⁵ stated that the amount of canine reduction that is often required to appropriately position these teeth aesthetically and functionally in three planes of space can be
excessive, and that the option of opening space and selecting a restorative solution must be considered. The edentulous sites require sufficient inter-radicular space in order to accommodate an implant. Therefore, adequate orthodontic preparation is essential in order to provide space for both the restoration and the implant. In cases of long-standing edentulous areas, bone grafting is often necessary to provide adequate thickness and height to the alveolar bone. A potential advantage of alveolar implant site development by distalising the canines that have erupted adjacent to the central incisors is that the alveolar bone in the lateral incisor site maintains adequate bone architecture with a lesser tendency to resorb. However, a long-term problem of implants in the aesthetic zone is the tendency for the attached gingiva to migrate apically, resulting in the implant neck becoming visible. This process was accelerated in Twin L., as illustrated in Figure 17.

The twin sisters were nearly in Class I molar relationships at the beginning of treatment. However, L. had a tendency to be slightly more Class II than her sister. Both had slight crowding in the lower arch, but extractions were not indicated. Furthermore, their flat facial profiles and obtuse nasolabial angles were contraindications to retraction of the upper anterior teeth, thereby eliminating space closure as an acceptable option. Accordingly, space opening with maxillary lateral incisor replacement was considered a better treatment option.

An interesting aspect of this case of identical twins is the slight but significant difference in outcomes, despite the identical treatment that they received.
The pre- and post-treatment tracing superimpositions in Figure 14 demonstrate a different degree of downward and backward mandibular rotation. In the case of M., the FMA increased by $+2^\circ$, compared with L., in whom the FMA decreased by $-2^\circ$. Given that identical twins share the same genetic code, any differences between their outcomes are attributable to environmental factors, which may relate to patient compliance. The slightly greater increase in lower face height seen in one twin may be due to her less rigorous compliance with instructions or in wearing elastics. It is possible that their orthodontic force systems may have differed with respect to the vertical component of force, resulting in a greater downward and backward rotation in one (M.).

An alternative possible explanation for this difference may be related to epigenetics, which postulates that there are additional factors at play, which act as a bridge between genetics and the environment. In a recent article, Williams et al. stated that two specific layers of information are encoded within the human genome. The first layer contains our genetic pattern encoded in the nucleotide sequence. The second layer of information contains an epigenetic code for development and maintenance that dictates when and where various genes are activated and deactivated during embryogenesis, growth, and throughout life. Therefore, an epigenetic role may be involved in the different outcomes seen in the twins.

Another important aspect that affects treatment outcome is patient compliance, not only during treatment but throughout the long-term retention period. The inferior compliance exhibited by Twin L. in the retention phase resulted in a marked degree of alignment relapse and a less aesthetic outcome.

A further consideration in the present study relates to the number of restorative options and retention appliances utilised by the twins. In treating edentulous sites in adolescents, definitive restorative options are delayed until the majority of jaw growth has finished, especially in the vertical dimension. The twins received several lateral incisor replacements, ranging from a removable appliance to a band-supported fixed appliance, to Maryland bridges and finally to implant supported crowns.

Figure 16. Final photographs after replacement of the maxillary lateral incisors by Maryland bridges at age 21.
The treatment of patients with congenitally missing maxillary lateral incisors presents a challenge to the clinician, especially when the canines erupt adjacent to the central incisors. The dilemma is whether to close the space by the substitution of the canines for the lateral incisors, or to open sufficient space for the replacement of the absent teeth. The latter option was selected for the twin girls presented.

1. By expanding the maxillary arch, laterally and antero-posteriorly, it was possible to create sufficient space to accommodate replacement lateral incisors.
2. This permitted the maintenance of the nasolabial angle by avoiding lip retraction during space closure.
3. The successful outcomes occlusally, functionally, and aesthetically may remain stable after the completion of treatment through a variety of restorative and retention appliances.
4. There are generally financial and biological costs involved in tooth replacement related to the restorative aspects (bridges, implant supported crowns) and surgical aspects (bone or soft tissue grafting).

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