Patient with asymmetric multiple hypodontia treated with autotransplantation of 2 premolars

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Tooth autotransplantation is performed in patients with congenitally missing teeth and those with traumatic tooth loss. We report a course of edgewise treatment of a girl with multiple congenitally missing teeth and residual features of ectodermal dysplasia, who was treated with autotransplantation of 2 premolars with developing roots. She was 8 years old at the beginning of the treatment. No signs of inflammation, root resorption, or pulp symptoms were observed during the 2.5 years of edgewise treatment after autotransplantation. Cervical external root resorption was detected 31 months postoperatively in 1 transplanted tooth (maxillary first premolar), and the same problem occurred in the other transplanted tooth (mandibular second premolar) 2 years later. Root canal treatment was immediately undertaken and resulted in inhibition of further pathology. Consequently, the survival of both transplanted teeth was achieved. The orthodontic treatment that included intervals related to diagnosis and treatment of root resorption of the transplanted premolars is described in detail. (Am J Orthod Dentofacial Orthop 2019;155:127-34)

Tooth autotransplantation is performed in patients with congenitally missing teeth and those with traumatic tooth loss. It is especially considered in patients with orthodontic indications for tooth extraction when potential donors are available.[1-4] In many cases, it is an attractive alternative to implants and prosthetic replacement of missing teeth.[5] Unerupted premolars are the most predictable donors.[6] According to most studies, the tooth should be transplanted at a proper stage of root development (1/2 to 3/4).[7] because this increases the possibility of pulp revascularization.[4] Normal healing after autotransplantation includes periodontal ligament restoration (radiologically accompanied by the formation of lamina dura and clinically by normal mobility and tooth eruption), pulp revascularization (associated with total or partial pulp obliteration), and further development of the root.[8]

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Many studies have proved that autotransplantation of premolars combined with orthodontic treatment can be the first treatment choice in patients with missing teeth when potential donors are available.[4,5,9,10] In 2002, Czochrowska et al[3] presented the results of a study of 28 patients. Only 3 of 33 transplanted teeth were lost after 9, 10, and 29 years postoperatively. The success criteria were not fulfilled for 4 other teeth because 2 transplants were ankylosed and another 2 had a crown-to-root ratio greater than 1, indicating a short root. No teeth were treated endodontically. The mean observation period was 26.4 years.

In 2004, Jonsson and Sigurdsson[9] published the study results of teeth autotransplanted with open or half-open apices. A 66% pulp survival was observed in developing teeth. Transplants with closed apices received endodontic therapy. Endodontic treatment resulted in the arrest of the inflammatory resorption in 2 teeth.

Ectodermal dysplasia was defined by Freire-Maia and Pinheiro.[11] According to the definition, it is a pathogenic development with an embryonic defect damaging the structures and tissues derived from the ectoderm. Oligodontia is 1 of the most characteristic features of this syndrome.

This case report describes the autotransplantation of 2 developing premolars in a patient with asymmetrical oligodontia and residual features of ectodermal

Table. Details of the treatment

Treatment duration/time after autotransplantation	*Treatment course*
June 2007: 2-year observation of potential donors' development	First appointment, extraoral and intraoral examinations, first panoramic radiograph showed oligodontia, diagnosis; orthodontic consultation for the treatment plan was agreed with the surgeon
October 2007	Patient received a maxillary removable appliance to distalize tooth #3
July 2009	Autotransplantation of 2 developing premolars: maxillary first and mandibular second from the right side to replace 2 missing teeth in the maxilla on the left side
March 2010/8 months	Bonding the fixed appliance in the maxillary arch excluding both transplanted premolars (#5 and #29), archwire, 0.012-in nickel-titanium
May 2010/10 months	Tooth #5 (mesial transplant) tied to archwire
July 2010/12 months	Changing the wire for 0.014-in nickel-titanium; transplanted #5 derotated after 11 weeks (palatal cusp moved distally in relation to the position of buccal cusp)
August 2010/13 months	Changing the wire for 0.018-in nickel-titanium; bonding the bracket on transplanted #29 (distal transplant)
October 2010/15 months	Nickel-titanium coil spring placed between #5 and #14 to mesialize #5 with 1-mm activation
March 2011/20 months	Next 1-mm activation of a coil spring to mesialize #5; tooth #29 drifted without any force
September 2011/26 months	Changing the place of the bracket on fully erupted #5; #6 was completely mesialized; 1-mm activation of the coil spring on #5
December 2011/29 months	Placing an elastic chain from #6 to #5; #5 was mesialized; the place of the coil spring changed from #5 to #29; the coil spring for mesialization of #5 was in use from 2010 to 2011 (14 months)
February 2012/31 months	1-mm activation of the coil spring on transplanted #29; panoramic radiograph showed cervical root resorption of donor #5, which was also confirmed on intraoral radiograph (22 months of donor's movement); deactivation of transplanted #5 from fixed appliance; transplanted #5 underwent endodontic treatment; activation of the wire to upright transplanted #29
April 2012/33 months	Change of the bracket position on transplanted #29 to derotate it; return to 0.012-in nickel-titanium wire
July 2012/36 months	Return to 18-in nickel-titaniumwire; open coil spring between teeth #14 and transplanted #29; intraoral radiograph of transplanted #5 confirmed that resorption did not increase
August 2012/37 months	Transplanted #29 contacted the mesial transplanted #5; tooth #5 retied to wire
October 2012/39 months	Bonding of the mandibular fixed appliance; Ulti-Mim .022-in, MBT, 0.012-in nickel-titanium wire
December 2012/41 months	Removal of the bracket from transplanted #5
June 2013/47 months	Composite buildups on teeth #8 and #9 performed
July 2013/48 months	Placement of nickel-titanium open-coil spring between teeth #6 and #8 to procline maxillary incisors
February 2014/55 months	Intraoral radiograph of transplanted #29 showed cervical root resorption; endodontic treatment of transplanted #29
May 2014/58 months	Conclusion of full fixed appliance treatment; removal of the fixed appliance
November 2015/64 months	Composite reconstruction of transplanted tooth #5

International tooth-numbering system: #3, maxillary right first molar; #5, maxillary right first premolar; #6, maxillary right canine; #7, maxillary right lateral incisor; #8, maxillary right central incisor; #9, maxillary left central incisor; #11, maxillary left canine; #12, maxillary left first premolar; #13, maxillary left second premolar; #29, mandibular right second premolar.

dysplasia. The orthodontic treatment, healing complications of the transplants, and their treatment are presented and discussed (Table).

DIAGNOSIS AND ETIOLOGY

In June 2007, an 8-year-old girl reported to the orthodontic private practice. The extraoral examination showed anterior rotation of the mandible, a concave profile, decreased lower anterior facial height, and a slight asymmetry with a left lateral shift of the mandible. The intraoral examination showed hypertrophy of the upper lip frenulum with diastema and displacement of the midline of the mandibular arch to the left side. According to the Angle classification, a cuspal Class II

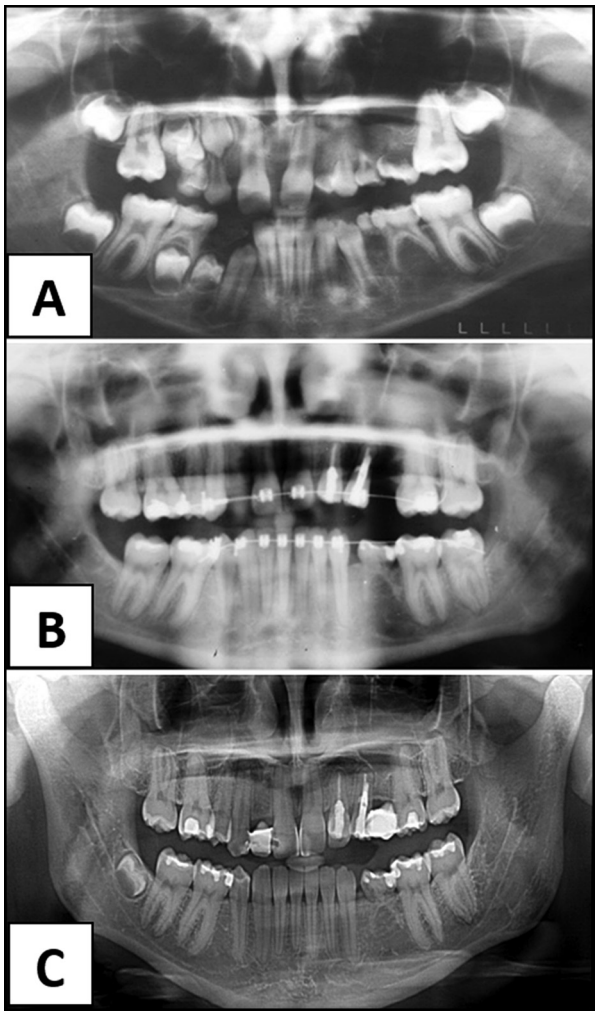

Fig 1. Panoramic radiographs: **A**, 2 years before tooth transplantation (June 2007); **B**, before the end of treatment; **C**, 18 month after orthodontic treatment (November 2015).

relationship was present on the right side with a full Class II relationship on the left side. The patient had previously lost her deciduous teeth, resulting in the partial lack of space for the maxillary right premolar and both mandibular first premolars. Family history showed hypodontia of both maxillary lateral incisors in the patient's mother and maternal grandmother. The patient had some features of ectodermal dysplasia, such as hypodontia, fair hair, flattened bridge of the nose, prominent forehead, lower anterior facial height reduction, and concave profile. However, hypotrichosis, nail dysplasia, and hypohidrosis were not observed. The panoramic radiograph showed oligodontia of both maxillary lateral incisors, the maxillary left canine and first and second premolars, and the mandibular left first and second premolars. Therefore, there were only 3 permanent teeth in the maxillary left quadrant: the central incisor and the first and second molars (Fig 1, *A*).

TREATMENT OBJECTIVES

Based on the diagnosis and after consultation with the patient's parents and the surgeon, the following objectives were developed: (1) autotransplantation of 1 maxillary and 1 mandibular developing premolar from the right side to the left side in the maxilla to replace missing teeth; (2) orthodontic alignment of the teeth (including the transplanted premolars); (3) closing the spaces at the donor sites; (4) reducing the space between the mandibular left first molar and canine; (5) maintaining the mandibular deciduous left second molar as long as possible to retain bone for the future implant or transplant in this area; and (6) achieving a functional occlusion in these dental conditions.

TREATMENT ALTERNATIVES

In an adult patient, implants replacement at the sites of the missing maxillary left canine and first and second premolars could be an alternative. However, due to the patient's young age, implants were contraindicated. A conventional prosthetic bridge was ruled out because of the extensive edentulous area. Microimplants could have been used to improve the mesial movement of maxillary and mandibular dentitions, but the patient's parents declined this treatment option.

TREATMENT PROGRESS

In March 2009, after obtaining the succeeding x-rays—orthopantomogram, dental radiographs (Fig 2, *A* and *B*), and tomography (Fig 3, *A*)—and after consultation with an oral surgeon, the date of autotransplantation surgery was set. The developing maxillary right first premolar and mandibular right second premolar were to be the donors. The decision was made to extract the maxillary deciduous left lateral incisor and second molar and both mandibular first molars because of advanced root resorption.

The maxillary removable orthodontic appliance (with the screw to achieve distalization of the maxillary right first molar) was designed to facilitate access to the unerupted developing maxillary right first premolar, which was scheduled for autotransplatation.

SURGICAL PROCEDURE

The surgery was performed under local anesthesia in July 2009. Full-thickness flaps were raised in the right sides of the mandible and the maxilla to enable access

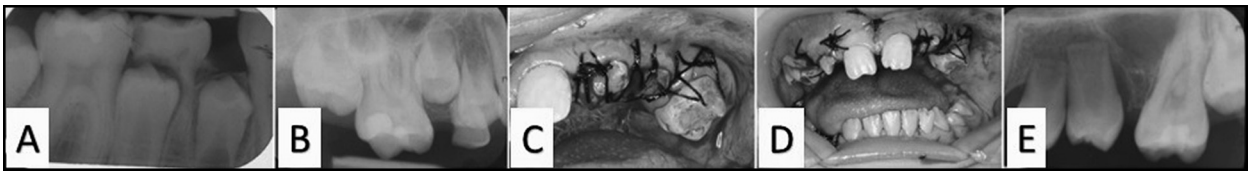

Fig 2. Two premolar donors before and immediately after teeth transplantation: **A**, mandibular right second premolar before transplantation; **B**, maxillary right first premolar before transplantation; **C**, operating field: 2 donors (maxillary right first premolar and mandibular right second premolar) after surgery; **D**, intraoral view after surgery; **E**, intraoral radiograph immediately after surgery (July 2009).

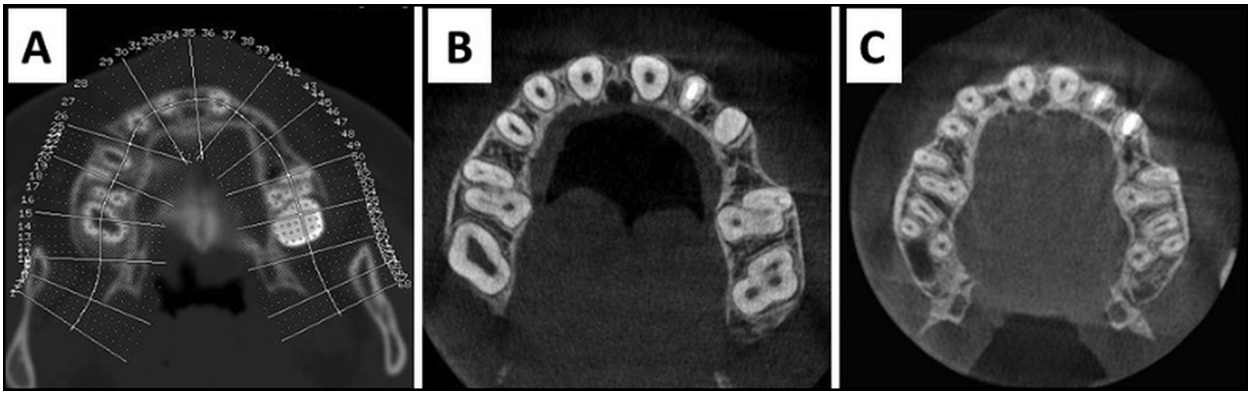

Fig 3. Comparison of the sagittal bone levels during orthodontic treatment: **A**, before autotransplantation; **B**, after endodontic treatment of the maxillary right first premolar; **C**, normal development of the alveolar bone after transplantation of premolars to the left side of the maxilla. The alveolar bone maintained its width during orthodontic treatment.

to the unerupted donor teeth (maxillary right first premolar and mandibular right second premolar). The buccal plates were carefully removed to prevent damage to the periodontal ligament of the developing roots of the donors. Then the deciduous canine and molar were extracted. Separate artificial sockets were prepared in the maxilla between the left central incisor and left first molar. Osteotomies were performed with surgical burs. The sockets were adequate to accommodate the roots of the premolars with an additional 1-mm space on each side of the transplants. The donor teeth were gently removed and transplanted to the artificial sockets. Both teeth were slightly rotated to fit the deficient palatobuccal dimensions of the alveolar process. The donors were stabilized in semierupted positions using only sutures (Fig 2, *C* and *D*). The dental radiograph was taken to check the result of the surgery (Fig 2, *E*). The patient received 1 intramuscular corticosteroid injection (8 mg, dexamethasone), 500 mg of amoxicillin (3 times daily for 7 days), and 200 mg of ibuprofen (3 times daily for 3 days after the surgery). Healing was uneventful with no signs of bleeding or edema.

Follow-up appointments were scheduled at 1 month (for suture removal) and then at 3, 6, and 12 months and annually after transplantation. No signs of pathology regarding soft or hard tissue healing were detected up to 2 years after the surgery. The pulp of the transplanted teeth underwent partial or complete obliteration and responded to thermal and electrical stimuli within normal limits. Clinical examinations showed normal periodontal tissues at the transplanted and adjacent teeth. Both transplants had normal mobility (stage 1 assessed according to Mühlemann's classification[12]) and normal eruption to the occlusal contacts.

PROGRESS OF ORTHODONTIC TREATMENT

In March 2010, 8 months after transplantation the maxillary right first premolar (mesial transplant) and the mandibular right second premolar (distal transplant) (Fig 4, *A*), orthodontic treatment with fixed appliances began. A maxillary fixed appliance (Ulti-Mim, MBT system, 0.022-in; Ortho Classic, McMinnville, Ore) was bonded to the maxillary teeth excluding the

Fig 4. Intraoral changes in position of 2 donor teeth during orthodontic treatment: **A**, after an 8-month follow-up of the development of the transplanted teeth (March 2010); **B**, transplanted teeth tied to the archwire (August 2010); **C**, mesialization of the maxillary right first premolar with the nickel-titanium open-coil spring; **D**, disconnection of the maxillary right first premolar from the arch; **E**, after mesialization of the mandibular right second premolar in place of the maxillary left canine; **F**, both fixed appliances were removed, and an adhesive fiberglass-reinforced bridge was in place on the maxillary right lateral incisor (May 2014); **G**, cosmetic composite bridges in the area of the maxillary left canine and first and second premolars (November 2015).

transplanted premolars. Two months later, glass ionomer temporary splints (Ketac-cem; 3M Unitek, Monrovia, Calif) on the mandibular molars enabled full-time disarticulation of the bite. The mesial transplant was tied to the 0.012-in nickel-titanium archwire. Two months later, it was tied to a 0.014-in nickel-titanium archwire. Subsequent intraoral x-rays showed normal development of both autotransplanted teeth (Fig 5, *A*).

In August 2010, a bracket was bonded to the distal transplant and the 0.014-in nickel-titanium archwire was changed to an 0.018-in nickel-titanium archwire (Fig 4, *B*). After the leveling and alignment phase, further treatment consisted of gentle mesialization of the mesial transplant by a nickel-titanium coil spring (light open-coil spring; GAC International, York, Pa) activated 1 mm per month; this lasted for 14 months (Fig 4, *C*). The distal transplant spontaneously moved mesially without any active elements. Its further mesialization by the light nickel-titanium open-coil spring started in December 2011 and lasted 4 months. After that, the palatal cusps of the transplants were ground to exclude them from the occlusion.

In February 2012 (31 months after transplantation), after mesialization of the maxillary right first premolar to replace the missing lateral incisor, cervical external root resorption was observed at the mesial root surface of this tooth (Fig 5, *B*). The patient was referred for endodontic treatment, which was completed in the same month. The decision was made to exclude the affected transplanted tooth from the arch and to continue mesialization of the distal transplant to replace the missing canine (Fig 4, *D*). The dental radiograph and computed tomography, performed after 3 months, showed no signs of progressive resorption of the mesial transplant. No signs of pathology of the distal transplant were seen (Figs 3, *B*, and 5, *C*). In August 2012, the mesial transplant was retied to the archwire; 2 months later, the mandibular fixed appliance was placed after full eruption of the mandibular premolars. The intraoral radiograph taken after 2 months showed slightly greater resorption in the mesial transplant but no pathologic changes in the distal transplant. After consultation with the surgeon, an orthodontic bracket was removed from the mesial transplant (Fig 4, *E*).

Composite widening of both maxillary central incisors was performed, and the space maintaining the area corresponding to the missing maxillary right lateral incisor was planned to increase inclination of the anterior teeth to improve the concave profile.

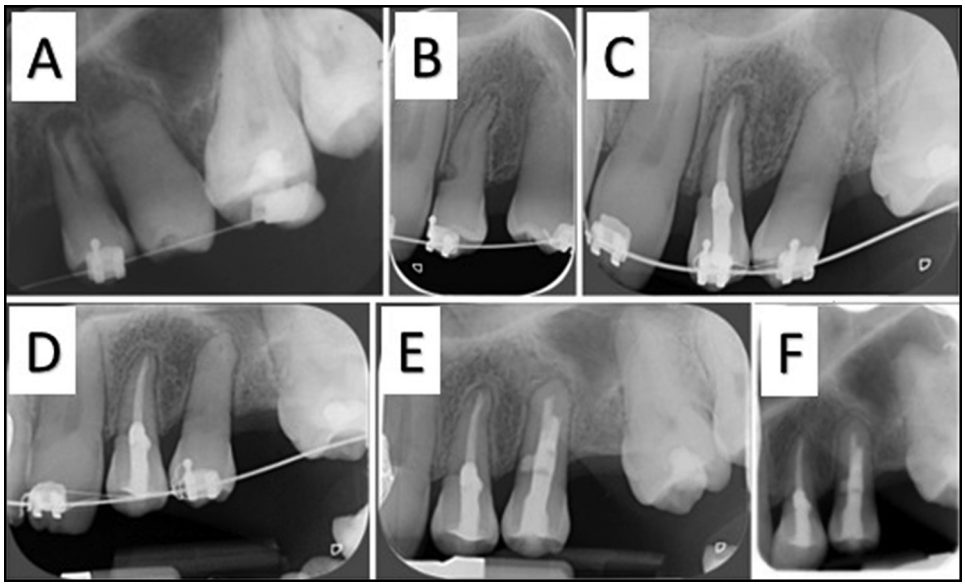

Fig 5. Radiographic presentation of the sequel of the 2 donors' healing, complications, endodontic treatment, and stability during and after orthodontic movement: **A**, maxillary right first premolar tied to the 0.012-in nickel-titanium archwire (10 months after transplantation); **B**, cervical external root resorption on the mesial root surface of the maxillary right first premolar (31 months after transplantation); **C**, endodontic treatment completed, with no signs of resorption of the mandibular right second premolar (36 months after transplantation); **D**, cervical external root resorption on the mesial root surface of the mandibular right second premolar (55 months after transplantation); **E**, endodontic treatment of the mandibular right second premolar completed; **F**, no relapse of the maxillary right first premolar and mandibular right second premolar root resorption (November 2015).

In February 2014 (55 months after transplantation), the intraoral radiographs showed external root resorption of the distal transplant, and the patient was referred for endodontic treatment (Fig 5, *D* and *E*).

In May 2014, the fixed appliance was removed (Fig 1, *B*). An adhesive fiberglass-reinforced bridge was used to replace the missing maxillary right lateral incisor (Fig 4, *F*), and an Essix retainer secured the space between the distal transplant and the first molar for future prosthetic replacement. After a 21-month follow-up and no signs of progressive resorption in either of the transplanted teeth (Figs 3, *C*, and 5, *F*), a cosmetic composite bridge was bonded on the distal transplant and the first molar (maxillary left first molar and mandibular right second premolar respectively) (Figs 1, *C*, and 4, *G*). The patient remains under our supervision and has follow-up appointments every 3 months.

DISCUSSION

A number of studies have emphasized that autotransplantation can be an effective treatment.[3,13,14] Czochrowska et al[3] found a survival rate of 90% with good long-term stability, over a mean observation period of 26.4 years. However, the risk of complications should be considered when the surgical procedure is planned. The most common complications include ankylosis and inflammatory resorption.[13,14] Both processes belong to the classification of external resorption by Ne et al,[15] based on the clinical symptoms and the histopathologic picture. The frequency of ankylosis is about 4.3% to 12%.[9]

The advantage of autotransplantation is the ability of a developing donor tooth to induce alveolar bone growth.[15-18] This is not the case for removable dentures resulting in the loss of alveolar bone that worsens the conditions for possible future implantation. Autotransplantation supports bone regeneration; therefore, even when the transplant is lost, the normal alveolar process is better prepared for a dental implant.[19]

In 2007, studies were presented that compared donor teeth classified into 3 stages of root development. It was proved that autotransplantation of a tooth with closed apices does not lead to external resorption. The pulp survival of transplants was 60.7%.[20] A similar outcome was also found by Choi et al.[21]

Autotransplantation creates an opportunity to improve the occlusion and maintain good facial esthetics in patients with cleft palate.[22] In addition, this kind of treatment may induce alveolar bone stimulation and prevent atrophy of the grafted bone.[23] Tooth transplantations to bone-grafted clefts have been reported to give stable results.[23,24]

In our patient, the treatment plan included transplantation of the 2 developing premolars from the unaffected (right side of the mandible) and less affected by hypodontia (right side of the maxilla) sites to the area with 4 missing permanent teeth (left side of the maxilla). It was the proper time to begin the orthodontic treatment, which prepared the space for the autotransplantations. The stage of root development of the premolars was adequate to promote revascularization.[25-27] The transplants would also initiate further development of the maxillary alveolar process on the left side.

Root resorption is a rare complication of tooth transplantation. Invasive cervical root resorption, which occurred in our patient, is an uncommon and aggressive form of external root resorption.[28] Typically, it begins at a focal point on the root surface below the epithelial attachment in the cervical area. It progresses coronally, apically, and circumpulpally into the dentin, halting only at the predentin layer surrounding the pulp, and destroying dental hard tissues asymptomatically.[29] According to Trope,[30] 2 conditions must be met for the resorption to progress: the loss or alteration of the protective layer (precementum or predentin) and inflammation near the unprotected root surface. Resorption can appear long after surgery. Its type can be confirmed only by a biopsy, where the resorption lacunae on the root surface and cavities in the dentin can be seen.[31] Removal of the affected hard tissues followed by curettage of the granulation tissue and restoration of the defect with either glass ionomer cement or composite resin is 1 method proposed to stop the resorption process.[32,33] However, Scandinavian authors have recommend endodontic treatment as the only necessary treatment option.[9] This type of treatment was performed, and it was successful in our patient.

In our patient, invasive cervical root resorption affected the maxillary right first premolar 31 months after transplantation (23 months after beginning the orthodontic treatment) and the mandibular right second premolar 55 months after transplantation (47 months of the orthodontic treatment). The transplants were positioned as close as possible to the incisors for the safety of their root surfaces during surgery. Resorption at the mesial sites of both transplanted premolars occurred during mesialization of the transplants to their final positions. According to a recent study, external cervical root resorption at the compression sites is associated with the amount of tooth movement.[34]

Both teeth were treated endodontically and controlled radiologically. After the initial increasing resorption, dental radiographs showed remission of pathology in both transplants.

The fiberglass-reinforced bridge replacing the missing maxillary second premolar on the left side was placed on the first molar and the mandibular right second premolar. This replacement may have increased the load on the transplant, but the patient's young age (16 years) prevented replacement of the second premolar with a dental implant. It was important to secure a space for a future implant using an adhesive fiberglass-reinforced bridge and the Essix retainer until the implant is placed in the area of the missing second premolar. The final type of prosthetic replacement of the maxillary second premolar was not defined in the initial treatment because in a patient with 4 teeth missing in the maxilla, transplantation was the treatment priority, and the remaining space at the left side of the maxilla could not be precisely predicted at that time. Additional replacements could be considered after alignment of the transplanted teeth and would depend on size of the remaining space, alveolar bone volume, relationship to the floor of the sinus, and the patient's age and preferences.

CONCLUSIONS

Autotransplantation of the 2 developing premolars enabled replacement of 2 of 4 missing teeth on the left side of the maxilla in a patient with asymmetrical oligodontia.

The development of the transplants' roots and their orthodontic movements improved the alveolar bone volume in the initially deficient area.

This treatment allowed establishing a stable and functional occlusion without the use of implants or a removable prosthesis. These are contraindicated in growing patients.

The thorough follow-up of the transplanted premolars enabled timely detection of complications so that treatment made it possible to preserve the transplanted teeth.

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