

Orthodontic Treatment, Including Transplanted Teeth and Dento-Alveolar Osteotomy, for Dental Ankylosis after Multidisciplinary Treatment for Rhabdomyosarcoma of the Cheek: A Case Report and Review

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Abstract

Multidisciplinary treatment for head and neck malignant tumors is often performed in children and adolescents. This treatment strategy may induce dental morphological aberrations and missing teeth, resulting in leading to problems with occlusions. A 14-year-old male underwent multidisciplinary treatment for rhabdomyosarcoma of his right cheek when he was three years old. He claimed a serious malocclusion. The hopeless teeth were extracted. Simultaneously, the bilateral lower first premolars were auto transplanted to the upper right canine and first premolar regions. Total orthodontic treatment was initiated six months after the surgery. Subsequently, a segmental osteotomy was performed because of dental ankylosis of the right upper central incisor. Finally, a good occlusion was created. Orthodontic treatment that includes the transplanted tooth after healing can create an ideal dental arch and good occlusion. If dental ankylosis is determined, a dentoalveolar osteotomy should be performed to create a good occlusion.

Key Words: Multidisciplinary treatment, Tooth transplantation, Rhabdomyosarcoma, Dento-alveolar osteotomy, Dental ankylosis

Introduction

Multidisciplinary treatment, which combines tumor resection, chemotherapy (CT), and radiotherapy (RT) for malignant tumors in the head and neck region, is often performed in children and adolescents [1-3]. The outcomes of this treatment strategy have been reliable; however, there are a number of disadvantages accompanying this treatment, such as the disturbance of growth and development in the head and neck region [4]. Furthermore, because the tooth is affected negatively by CT and RT during its development, dental morphological aberrations and missing teeth can result [5].

One of the treatment options for a missing tooth is auto-tooth transplantation. This method is a quite reliable option, and its long-term survival rate is more than 90% [6-10]. There are two essential factors for the success of auto-tooth transplantation. One factor is an intact periodontal ligament [11], and the other is an appropriate size of the recipient site [12]. The risk factors for the failure of auto-tooth transplantation are age (40 years or more) and using a molar tooth as a donor [5]. However, there is no report that has examined the survival rate of auto-tooth transplantation at the irradiated region. According to previous reports describing dental implantation at the irradiated region [13-16], auto-tooth transplantation may succeed at the irradiated region if the total irradiation dose is less than 45 Gy.

While the harmful effects of CT to the periodontium remain unclear, there are a number of reports that refer to the deleterious effects of RT to the periodontium [17,18]. In particular, the periodontal ligament has a high sensitivity to radiation. For instance, RT has been shown to expand the periodontal space [19] or to induce periodontal attachment loss, resulting in an increase in the occurrence of osteoradionecrosis [20]. However, there is no report that has referred to the relationship between CT or RT and dental ankylosis.

In this report, we describe a patient who had submitted to such a multidisciplinary treatment for rhabdomyosarcoma of his right cheek in childhood. For a dental morphological aberration and serious malocclusion, he underwent the extraction of the abnormal teeth and auto-tooth transplantation at the irradiated region. Subsequently, orthodontic treatment and dento-alveolar osteotomy for the dental ankylosis created a good occlusion.

Case report

This study received ethical approval from the institution. A 14-year old male visited the Department of Dentistry and Oral Surgery, University of Fukui Hospital, due to malocclusion (Figure 1). He had undergone a tumor resection when he was three years and had subsequently received CT (vincristine, actinomycin D, cyclophosphamide and cisplatin) and RT (Linac X-ray, total 44 Gy) for rhabdomyosarcoma of his right cheek, according to the Intergroup Rhabdomyosarcoma study-III protocol [2], one year after surgery. The patient has had no recurrences since submitting to the multidisciplinary therapy. According to routine examinations, including orthopantomograph (OPG) (Figure 2), cephalometric analysis (Table 1), and clinical findings, some problems had been observed, as listed below:

- the first molar occlusal relationship was class III on the right and class I on the left
- crowding of the bimaxillary anterior teeth due to a discrepancy between the size of the alveolar bone and the width of teeth
- remaining upper right deciduous cuspid
- labioinclination of the bimaxillary incisors
- short roots of the upper right lateral incisor, canine, premolars and the upper left lateral incisor
- relatively short roots of the upper left central incisor, second premolar and the upper right central incisor

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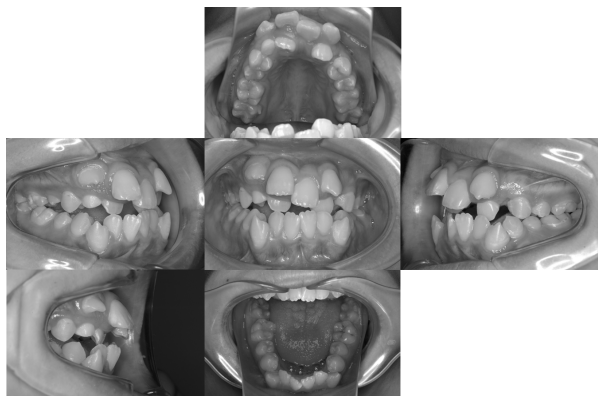


Figure 1. Intraoral findings at his first visit.



Figure 2. Orthopantomography at his first visit.

Table 1. Cephalometric analysis before and after treatment. T1; pre-treatment, T2; before corticotomy, T3; at retainer setting.

	Mean	SD	T1	T2	T3
Facial angle	85.07	5.76	87.3	87.7	87.9
Convexity	5.60	4.33	10.0	8.7	8.8
A-B plane	-5.10	3.28	-4.9	-3.7	-4.1
SNA	81.82	3.09	86.4	85.8	86.1
SNB	78.61	3.15	81.4	81.7	81.8
Mandibular Plane	26.25	6.34	30.3	29.3	29.2
Y-axis	65.71	3.27	62.3	61.9	62.0
Occlusal Plane	9.52	4.01	7.1	9.2	8.2
Interincisal	129.66	8.99	99.9	122.7	124.0
U-1 to FH plane	108.94	5.62	129.2	116.2	114.9
L-1 to Mandibular plane	94.67	7.21	100.6	91.9	91.9

Based on the problems mentioned above and the patient's wishes, a treatment procedure was planned. Because the reason for his malocclusion was a discrepancy between the size of the alveolar bone and the width of teeth, we extracted his upper right lateral incisor, deciduous cuspid, canine and upper right first premolar and upper left lateral incisor, which were considered to be hopeless because of disabilities in their root development. Subsequently, the bilateral lower first premolars were extracted and transplanted into the region of right upper canine and first premolar. These treatments

created the occlusion after that the bilateral upper lateral incisors and bilateral lower first premolars were lacking. Although the length of roots of the upper right central incisor, upper right second premolar and upper left central incisor were relatively short, these teeth were considered to be maintained. If the teeth had been lost, dental implants would have been inserted. Root canal fillings of the auto-transplanted teeth were performed after three weeks (*Figure 3*), and orthodontic treatment was initiated two months post-surgery.



Figure 3. Intraoral findings when the root canal fillings of transplanted teeth were performed.

After two years, adento-alveolar osteotomy was performed at the upper right central incisor due to its dental ankylosis (Figure 4). A retainer was set six months later (Figure 5). No

root resorption, including at the transplanted teeth, was observed on OPG (Figure 6).

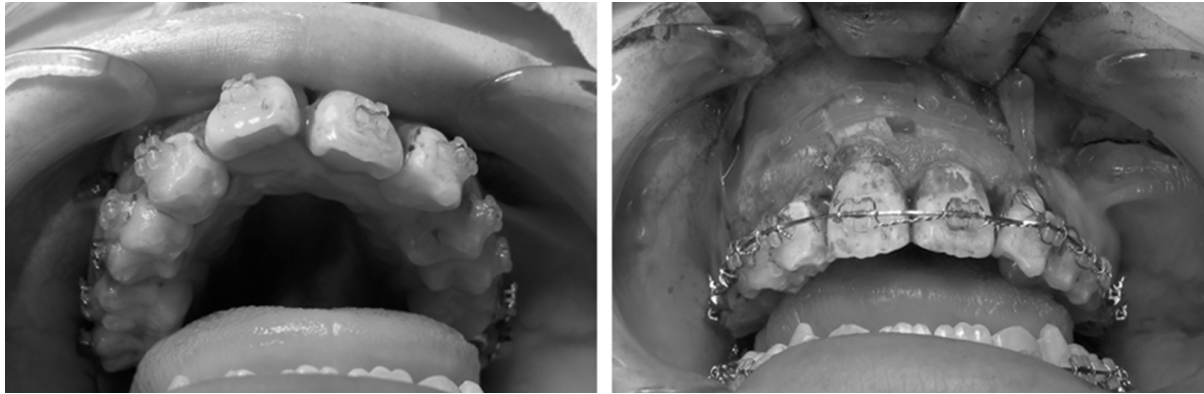


Figure 4. Intraoperative finding of the corticotomy before osteotomy (left) and after fixation with an absorbable plate (right). The bone particle was filled into the gap.



Figure 5. Intraoral findings at setting of a retainer.



Figure 6. Orthopantomography one year after the orthodontic treatment. No root absorptions of transplanted teeth were observed.

Discussion

It is well-known that both CT and RT adversely affect dental development during the incipient stages, and the combination of these two therapies increases the deleterious effects. The present patient underwent a systemic CT and local RT for his

rhabdomyosarcoma. While he had hypoplastic roots for some teeth in the irradiated region, there were no abnormal teeth outside of this region. This finding meant CT did not provide a sufficient side effect to create the abnormal teeth by itself; rather, RT alone or the combination of CT and RT may induce such abnormal teeth. There is no report that has clearly found how much irradiation is needed to produce such a dental morphological aberration. According to our case, 44 Gy may induce dental morphological aberrations when systemic CT is performed simultaneously.

This case demonstrated that it is possible to transplant an auto-tooth at the irradiated region when the irradiation dose is less than 44 Gy, even if systemic CT is simultaneously performed. It is also considered possible to perform orthodontic treatment for the transplanted tooth if the periodontal ligament regenerates and the bioactivity of the jaw bone is maintained. When performing tooth transplantation, it is important to confirm that the irradiated region of the jaw does not suffer from osteoradionecrosis. Marx et al. [19] suggested that RT damages the capillaries at the lesion, resulting in hypoxia due to the ischemic condition. Thus, osteoradionecrosis of the jaw (ORNJ) may be developed. ORNJ may occur when the irradiated dose is approximately greater than 50 Gy [15]. In this case, the facial appearance was symmetrical, and no bilateral developmental differences were observed at the maxilla according to lateral and frontal cephalogram. No canting of the maxilla was observed. Moreover, the development of the maxilla was not disturbed by RT, according to the normal development of the mandible outside of the RT field. Cephalometric analysis also implied non-disturbed bone growth (Table 1). Therefore, the patient's bone activity was considered to be unimpaired.

There is no report that has reviewed the success rate of auto-tooth transplantation at the irradiated region. To predict whether tooth transplantation is successful, the availability of the success rates of dental implants at the irradiated region is informative. Colella et al. [16] reported that there is no significant difference in the implant success rate within 45 Gy, and Dholam and Gurav [13] concluded that RT did not influence the implant treatment if the amount was less than 45

to 50 Gy. Moreover, Javed et al. [14] stated that osseointegration is not affected by 50 to 65 Gy. According to these previous reports, biological activity should be considered to be inhibited by more than a 45 Gy RT dose. Hence, auto-tooth transplantation was considered to have little possibility of succeeding in such cases. The preservation of the periodontal ligament is one of the essential factors to successful auto-tooth transplantation. In this case, the donor teeth were considered not to be affected by RT because the donor teeth existed outside the irradiation area. In fact, the donor teeth were normal, and the periodontal ligament spaces of these teeth were observed on OPG.

While Fiorentino et al. [21] initiated orthodontic treatment six months after auto-tooth transplantation, Ziegler et al. [22] initiated orthodontic treatment four weeks after auto-tooth transplantation. Day [5] advocated that orthodontic treatment should be initiated 4-6 months after tooth transplantation. On the other hand, Kim et al. [6] reported that the soft tissue healing was completed two to four months after auto-tooth transplantation. Although the timing of the initiation of orthodontic treatment after the auto-tooth transplantation varies based on the previous reports, it is considered that the movement of the transplanted tooth should be initiated six months after the auto-tooth transplantation, at which point the periodontal membrane regenerates and the alveolar bone heals completely [5]. We initiated post-surgical orthodontic treatment two months after transplantation, but the transplanted teeth were moved six months post-surgery.

Park et al. [23] transplanted the tooth into a space provided during orthodontic treatment based on previous predictions, creating a good occlusion. Moreover, dental implant treatment is often performed instead of auto-tooth transplantation in similar procedures. Because orthodontic treatment was planned in this case, auto-tooth transplantation was considered better than dental implants due to its movability. Orthodontic treatment, including the transplanted teeth, provided a good outcome in this case.

RT sometimes affects the periodontium adversely [19,20,24,25]. Most reports have referred to periodontal attachment loss and damage to the periodontal ligament. Although there is no report that has specifically referred to the relationship between CT or RT and dental ankylosis, the upper right central incisor in this case showed dental ankylosis. In fact, orthodontic treatment was unable to move the tooth. The tooth was considered to be affected by RT due to its short root. Because the tooth had not had any episode of periodontal disease after CT/RT, the dental ankylosis was considered to be caused by damage to the periodontal ligament due to RT/CT. However, it was unclear why only this tooth showed dental ankylosis at the irradiated region. Dento-alveolar osteotomy was indicated, and a segment was moved to an ideal position with an ideal direction according to the previous report [26]. No infected bone was observed during surgery (Figure 4). The ideal dental arch was completed after surgery (Figure 5).

Conclusion

RT and/or CT for tumors of the oral and maxillofacial region often induce abnormal events, such as missing teeth and

abnormal dental morphology. Moreover, a size discrepancy can occur between the dental arch and alveolar bone. Malocclusion can be observed in these cases, and orthodontic therapy may be needed. If there are hopeless teeth due to the side effects of RT/CT, these teeth are extracted and normal teeth are transplanted into the region. Orthodontic treatment, including the transplanted tooth, can create an ideal dental arch and good occlusion. If dental ankylosis accompanied with CT/RT is observed and the bioactivity of jaw bone is normal, dento-alveolar osteotomy is indicated for the creation of an ideal dental arch.

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