

CASE REPORT

Tooth transplantation in cherubism and incontinentia pigmenti: report of two unusual casesJ.L. Jensen^{1,2} & B. Album¹¹Department of Oral Surgery and Oral Medicine, Faculty of Dentistry, University of Oslo, Blindern²National Resource Centre for Oral Health in Rare Medical Conditions. Lovisenberg Diakonale Hospital, Oslo, Norway**Key words:**

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Abstract

In children, autotransplantation has been an accepted treatment alternative in the Scandinavian countries for decades in cases of agenesis or trauma. When working with children with rare syndromes, lack of teeth is often so pronounced that there are no teeth available for transplantation. Even so, sometimes there are retained teeth that can be transplanted!

Here we present two patients with rare syndromes in which a retained tooth was transplanted. In both cases, neither the donor tooth nor the recipient site was ideal for transplantation. The first case is a girl with incontinentia pigmenti, 15 years old at the time of transplantation, and the second case is a boy with severe cherubism, aged 9.5 years at the time of transplantation. In the first case, a second molar with multiple roots retained at a mesial angle to the first molar in the upper jaw was transplanted to an infected site. In the second case, a retained first premolar in the upper jaw was transplanted to a cystic lesion. In both patients, the profit in the event of a successful transplantation was so great, that an attempt was made in spite of the negative circumstances. Both teeth were successfully transplanted and have been followed for more than 2 years with no ankylosis or pulp necrosis developing. We conclude that in a child with multiple missing teeth, the conventional rules of transplantation may be stretched.

Clinical implications

Transplantation of teeth, when successful, offers many advantages to dental implants and should be offered as a treatment alternative in all children.

Introduction

Transplantation of teeth to replace missing teeth is an alternative to orthodontic or prosthetic treatment, while implant treatment usually is performed at a later age. As opposed to treatment with implants, transplantation of teeth does not regularly result in ankylosis. Therefore, transplanted teeth will grow with the alveolar bone of the adolescent and infraocclusion does not occur in most cases. Transplantation employing single rooted premolars as the donor tooth is the most

common. The method was developed over 40 years ago by Slagsvold and Bjercke¹. They showed that root development continues after transplantation of teeth with unfinished root formation. The teeth maintain their capacity for functional adaptation and endodontic treatment is usually not necessary. In fact, Czochrowska *et al.*² examined 30 teeth in 25 of their patients and found a survival rate of 90% with a mean observation time of 26 years!

The optimal time for transplantation of premolars is when the root development has reached three quarters of the final root length thus ensuring further root growth and a vital pulp³. The prognosis for complete periodontal healing at this stage of root development is better than 90%. Transplantation of teeth most often takes place when there is crowding in one jaw and agenesis in the opposite jaw, or when upper front

teeth are lost due to trauma. The transplanted tooth keeps the bone in place and prevents early implant treatment⁴.

The transplantation procedure is technically difficult and a successful outcome requires that the donor tooth can be removed easily, without destroying the dental follicle. Furthermore, the presence of adequate space at the recipient site is mandatory, and mobility during the 2-week fixation period must be avoided. Thus, the transplant is placed in a half-erupted position using conventional sutures as opposed to rigid fixation. Periodontal healing is complete within 8 weeks⁵.

Lack of pulp canal revascularisation may occur, although this outcome is rare when the tooth has a wide-open apex⁶. Pulp revascularisation has been reported in 100% of the teeth with initial to half root development but decreased to 0% for teeth with fully developed roots³. This complication can be corrected by conventional endodontic procedures. Damage to the periodontal ligament must be avoided because it might lead to ankylosis, a more serious complication⁷. This complication prevents eruption and leads to a gradual substitution of the root with bone. Unfortunately, once developed, ankylosis cannot be stopped.

When the root of a transplanted premolar continues to develop and a normal periodontal ligament is established, such teeth can be moved by applying orthodontic forces like any other tooth which has erupted into occlusion⁴. An observation period of 6 months before orthodontic treatment is started is generally recommended.

Case 1

Incontinentia pigmenti (IP⁸) is a genetic disorder which affects skin, hair, teeth and nails. It is a subgroup of ectodermal dysplasia and is characterised by skin lesions and pigmentation in addition to the traditional findings of ectodermal dysplasia (defective development of hair, skin, nails, teeth and sweat glands). Discoloured skin is caused by excessive deposits of melanin. Most newborns with IP will develop discoloured skin within the first 2 weeks. The pigmentation involves the trunk and extremities. Tooth agenesis is a common finding, and teeth may be narrow and peg shaped.

Our patient, a female born in 1992, had a total of 10 teeth missing. At the time of treatment, in December 2007, she was bothered with itching, eczema and uneasiness due to her IP. The teeth 16, 55, 14–24 and 65 were present in the upper jaw. 17 was retained, and there was a wide median diastema. In the lower jaw, 36, 75, 34–44, 85 and 46 were present. All teeth were



Figure 1 Case 1, girl with IP, 15 years old at the time of transplantation, orthopantomogram (OPG) before transplantation.

smaller than usual. 12 had a very short root and had to be sacrificed. 65 was still in place, but had a deep filling and apical periodontitis with a fistula and advanced bone loss, hence the tooth had to be removed (Fig. 1).

Treatment

As 17 was retained and had no antagonist, the plan was to transplant 17 and remove 65 at the same time (Fig. 1). The girl was given antibiotics for a week prior to the transplantation. On X-rays, the roots of 17 appeared to be much less developed than the ideal 3/4. In this case, the short root was judged an advantage as a longer root of a molar would have complicated the transplantation.

Due to the position of 17, retained at a mesial angle to 16, it was hard to remove 17 carefully, without disrupting the dental follicle. In addition, when removing the tooth from the donor site, it became apparent that the roots were more developed than expected and that the diameter of the root was wider than the recipient site. In fact, after removal of 65 and careful curettage, the recipient site had to be enlarged twice to allow the placement of the donor tooth.

Follow-up

The anticipation of a successful result was limited due to the problems occurring during surgery. In addition, the orthodontist put a bracket on the tooth already after 4 months, but the actual forces employed were limited. In spite of all these negative events, the tooth is still in place with no signs of ankylosis, Figures 2–3.

Case 2

Cherubism is a rare genetic disorder characterised by bilateral, symmetrical enlargement of the mandible or

both the mandible and the maxilla, resulting from multilocular cysts composed of fibrotic stromal cells and osteoclast-like cells⁹. The phenotype ranges from no clinical manifestations to severe overgrowth of the mandible and maxilla. Onset usually is between ages two and five. The jaw lesions progress until puberty when they stabilise and then regress in most cases. Dental abnormalities include congenitally missing teeth, root resorption and displacement of permanent teeth by the jaw lesions.

Our patient is a boy born in 1998 with cherubism affecting both jaws, a very severe case. He had an anterior open bite with a relatively big horizontal overjet, and his maxillary midline was almost 6 mm to the left. Only teeth 16/46 and 25/36 were in occlusion. In the maxilla, teeth 16, 54, 53, 12, 11, 21, 63, 64, 25 and 26 were present, while 17, 15, 14, 13, 22, 24 and 23 had not yet erupted. 53 and 64 were mobile. 14 was retained in a horizontal position with the crown in a distal direction, and 23 and 24 were retained in transpositions (Fig. 4). In the lower jaw, 36, 32, 41, 42 and 46 were erupted while 33 and 43 were horizontally retained, and there was a small retained 37. All other teeth were absent (Fig. 4). The cherubism had resulted in root resorption and displaced and missing teeth in both jaws.



Figure 2 Case 1, OPG 14 days after transplantation of 17 to the region of 25.

Treatment

In the upper jaw, there was no room for 14. We decided to transplant 14 to the lower front region (Fig. 5a) even though a cyst related to his syndrome was present in this location. Also, the root of 14 was much shorter than the ideal length for transplantation as it was around 1/2 of the anticipated root length. However, there was no time to wait for additional root growth as 14 prevented the eruption of 15. Upon transplantation, 14 was easy to remove from the donor site due to its short root (Fig. 5b). The recipient site was prepared according to the usual routine, but after penetrating the alveolar cortical bone, a huge bony defect was found so that 14 was placed in a cavity rather than a prepared alveolus. The tooth was sutured in place according to routine procedure (Fig. 5c).

Follow-up

At review up to 2 years, the tooth is in place in a somewhat inferior position, but with a normal percussion sound not indicating ankylosis. The root is still short (Figs 6–8).

Discussion

In both patients, parts of their dental problems have been solved through transplantation of a tooth which would otherwise have been removed. The teeth still function more than 2 years later, even though the root length of both transplanted teeth were too short in both cases, one tooth was transplanted to a previously infected area, and the other to a cystic lesion. In most cases of transplantation performed at early stages of root development, a reduction in the final root length is seen¹⁰. Recently, a report on a case where two teeth were successfully transplanted to infected areas has been published¹¹.

Since the infected 65 had to be removed at age 15 in case 1, an implant could not be installed due to lack of bone and her young age, possibly implying future inposition of the implant. Without a transplanted

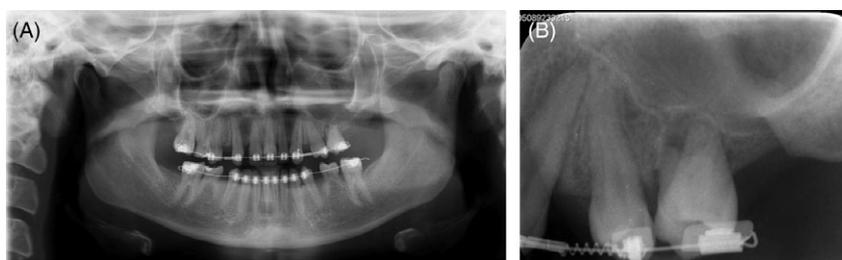


Figure 3 Case 1, 2 years after transplantation of 17 to the region of 25. OPG (A); X-ray (B).



Figure 4 Case 2, boy with cherubism, 9.5 years old at the time of transplantation. OPG before transplantation.

tooth in place, the alveolar ridge most certainly would have narrowed. Also, as the maxillary sinus was fairly extensive, later implant installation may have required bone transplantation. As demonstrated by Paulsen and Andreasen¹², a transplanted tooth creates bone in the alveolar process together with the eruption process. Thus, if this tooth fails in the future, implant treatment most probably can be carried out without prior bone transplantation.

For the boy with cherubism, only one of his many problems is solved through transplantation of one tooth. He has to undergo extensive corrective and reconstructive treatment after puberty due to his severe cherubism. As he stands the risk of losing most of the teeth in the lower jaw, one more tooth is always

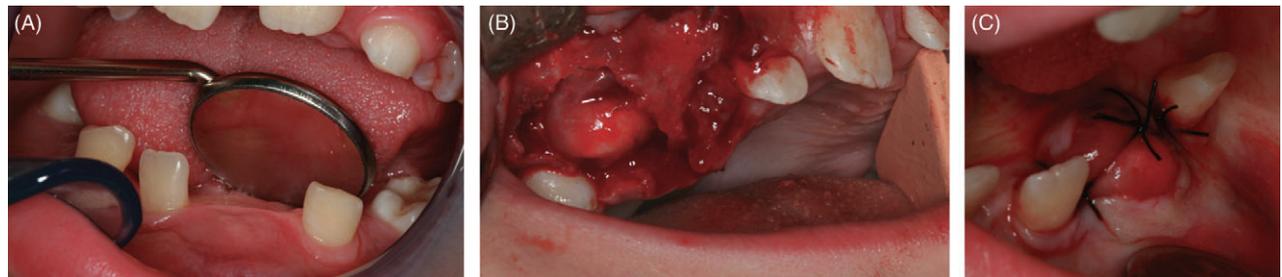


Figure 5 Case 2, clinical photos during the transplantation demonstrating the recipient site (A), the exposed donor tooth 14 with intact follicle (B) and the transplanted tooth fixed with sutures (C).



Figure 6 Case 2, OPG 14 days after transplantation of 14 to the lower front.



Figure 7 Case 2, clinical photos 7 months after transplantation of 14 to the lower front.



Figure 8 Case 2, OPG 1 year and 7 months after transplantation of 14 to the lower front.

an advantage. If nothing else, it can be used to support a partial denture until future implant installation.

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