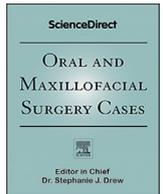




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# Plate-guided transport distraction osteogenesis of mandible after the excision of an ameloblastoma with inferior alveolar nerve preservation and dental implant treatment: A case report with long-term follow-up

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## ABSTRACT

Recently, the plate-guided transport distraction device (PGTD) is used as a reconstruction plate placed to fill the mandibular continuity defect. Additionally, when the mandible segments heal, osseointegrated implants (OI) with the dental prosthesis are recommended for the completion of the restoration, which is currently the gold standard. A 37-year-old female patient referred to our clinic with a complaint of pain on the right mandible. A partial mandibulectomy was performed including the ramus, angulus and corpus regions of the mandible extending from 1 cm anterior of the lesion to the subcondyle while the Inferior Alveolar Nerve (IAN) was preserved. A transport segment was created in a diameter of approximately 2 cm, including the first premolar, and a plate guided transport distractor was installed. One year after the resection and reconstruction surgery, dental implants were placed for prosthetic rehabilitation. A method of preservation of the IAN can be applied. After the radical resection of ameloblastomas, surgical management can be performed via transport distraction osteogenesis, and it has some advantages over microvascular free tissue transfer. Osseointegrated dental implants with the dental prosthesis are recommended to finalize the restoration after the healing of the mandible segments.

## 1. Introduction

Ameloblastoma is a true neoplasm of odontogenic epithelial origin, which usually occurs in the mandible and particularly in the angular region [1]. It can lead to cortical bone expansion, perforation, and invasion to surrounding soft tissues with locally aggressive behavior, high recurrence rate, and malignant potential. In general, ameloblastomas do not metastasize but the majority of ameloblastomas are commonly locally aggressive and disfiguring. Besides, they cause problems such as pain, asymmetry, and difficulty in speech and articulation [2].

The surgical treatment of ameloblastomas of mandibles is still controversial. Enucleation and curettage are conservative treatment approaches that have high recurrence rates, requiring repeated procedures [2–4]. On the contrary, radical excisions, including marginal or segmental mandibulectomies, present the challenge of reconstruction while related to much lower recurrence rates [2,5]. Marginal or

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segmental mandibulectomies are recommended for treating extensive cases which then lead to major deformities [6].

In addition, the difficult decision to be made is whether to preserve the Inferior Alveolar Nerve (IAN) or not. Generally, preservation of the nerve is attempted only if it lies outside the area of the tumor. However, the surgical removal of the neurovascular bundle is recommended when it is surrounded by a solid form of the tumor [7]. Mandible resections with nerve preservation for the treatment of cystic ameloblastoma were described by Becker though there was no evidence that the tumor invaded the nerve bundle [7,8].

Transport distraction osteogenesis (TDO) may be the treatment of choice for both hard and soft tissue reconstructions for ameloblastomas, to complete the restoration of form and function, dental rehabilitation is also required due to the loss of dentition. Currently, the plate-guided transport distraction device (PGTD) is used as a reconstruction plate which is placed to fill the mandibular continuity defect. This plate can be contoured before the resection and then reapplied to reconstruct the mandible by surgeons. The PGTD then follows the shape of the plate during distraction intimately so this provides 3-dimensional vector control during the distraction process [9].

Additionally, when the mandible segments heal, osseointegrated implants (OI) with the dental prosthesis are recommended for the completion of the restoration, which is currently the gold standard for providing oral and occlusal functions [2].

In the presented case, we attempted the radical excision of the ameloblastoma of the female patient's right mandible with the preservation of the Inferior Alveolar Nerve (IAN) simultaneously performing PGTD procedure, and rehabilitation with dental implants was carried out after the segment's healing by transport distraction osteogenesis by long-term follow up.

## 2. Case report

A 37-year-old female patient referred to our clinic (Erciyes University, Faculty of Dentistry, Oral and Maxillofacial Surgery Department, Kayseri, Turkey) with a complaint of pain on the right mandible in October 2011. She had been operated seven years ago for the treatment of a cystic lesion at the right posterior mandible. In the intraoral examination, a slight swelling was observed on the buccal and lingual aspects of posterior alveoli, and the patient had partial paraesthesia on the right lower lip. Panoramic radiography demonstrated a large multilocular radiolucency from right premolar teeth to coronoid process (Fig. 1). Computed tomography revealed a central radiolucent multilocular lesion sized  $5,5 \times 2,7 \times 2 \text{ cm}^3$  with thin cortical bone walls and destruction on the vestibule and lingual sides. Additionally line of IAN was located via CT (Fig. 2). A biopsy was performed under local anesthesia, and the histologic diagnosis was concluded Plexiform Ameloblastoma by the oral pathologist in certificated private pathology laboratory (Gören Histology & Pathology Laboratory, Ankara, Turkey).

Before surgery, the stereolithographic 3-D model was taken by CT scan for designating distraction borders, and reconstruction plate was precounted. The submandibular extraoral incision was conducted, and then exposing the tumor intraoperatively. The reconstruction plate was adapted, and the distraction lines and the borders were drilled in the healthy bone. A partial mandibulectomy was performed including the ramus, angulus and corpus regions of the mandible extending from 1 cm anterior of the lesion to the subcondyle, while the IAN was preserved using piezzo surgery and pulling through thanks to blunt separator devices carefully. A transport segment was created in a diameter of approximately 2 cm, including the first premolar, and a plate guided transport distractor (KLS Martin Group, Tuttlingen, Germany) was installed in single stage (Fig. 3). After a latency period of nearly 9 days, the distractor was activated twice a day at a rate of 1 mm/day. This activation was continued for 21 days until the transport segment could not be moved further. Five months later, approximately 25 mm new bone segment was occurred by the distraction device, and then it was removed, leaving the reconstruction plate in place. The anesthesia on the right lower lip and chin area was resolved three months after the operation.

One year after the resection and reconstruction surgery, dental implants were placed for prosthetic rehabilitation. However, the implant that was placed in the newly formed bone in the distraction gap was lost one month later, and then a second implant was placed after one-year follow-up. Dental implant (Tissue Level, Straumann, Switzerland) supported fixed prosthetics were restored after the completion of the osteointegration process (Fig. 4). Consequently, the oral and occlusal function was provided, and the patient was referred to the 6 months controls and then for 1-year follow-up (Fig. 5).

Approximately three years after the resection procedure, the patient referred to our clinic with a pain in the posterior mandible, and also complained about the mobilization of the reconstruction plate (Figs. 6 and 7). Therefore, the plate was removed, and no further reconstructions were performed. Also, we have seen that the optimum fibrous healing happened which filled the gap between segments. No recurrence of the tumor was detected on the cone beam computed tomography sections, and also there is no pathological finding in



Fig. 1. Preoperative panoramic radiograph of the patient with ameloblastoma at the right mandible (arrows).



Fig. 2. Destruction on the lateral (a) and lingual (b) sides of the mandible, axial view shows lingual bone destruction (c).



Fig. 3. (a) Preservation of inferior alveolar nerve (arrow) during resection. (b) Plate-guided transport distraction osteogenesis device.

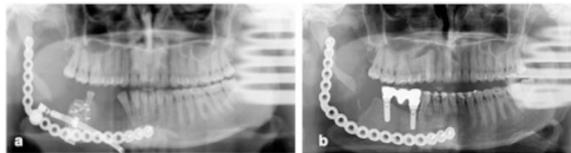


Fig. 4. (a) The transport segment (arrow) and distraction gap. (b) Restoration of the edentulous bone with an implant-supported fixed prosthesis after orthodontic correction of canine tooth.



Fig. 5. (a,b) Intraoral view of patient with provided oral and occlusal functions via dental implant supported prosthesis.

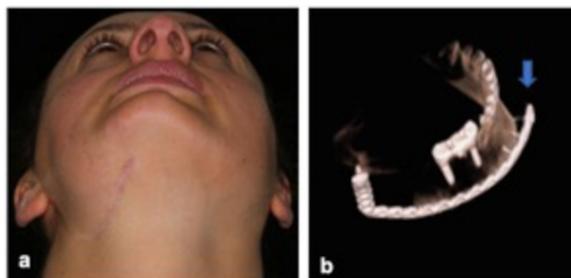


Fig. 6. (a) Deformed chin due to the loose end of the reconstruction plate. (b) 3D CT shows that the plate had lost connection with bone.

the recent follow-up panoramic radiographic view (Fig. 8). Additionally, the patient reported being satisfied with her appearance and (Fig. 9).

### 3. Discussion

The management of ameloblastomas is affected by factors like the patient's age, systemic disease, the location and size of the tumor,



Fig. 7. Cross-sectional views of the reconstruction plate on the condyle (a), and on the chin (b).



Fig. 8. There is no pathological finding in the follow-up panoramic radiographic view.

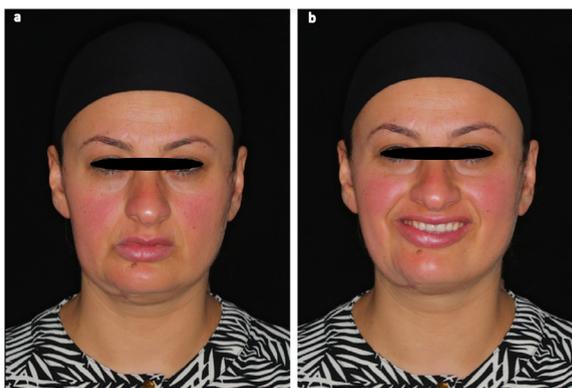


Fig. 9. (a,b) Recently front views of patient's face.

the radiographic and histologic features of the tumor, and the experience of the surgeons [7]. The decision should be concluded histologically whether the Ameloblastoma is aggressive or not with an incisional biopsy because it may be representative of the histological subtype [10] Radical excision with a safety margin of 1 cm is the accepted standard of treatment modality in certain types of Ameloblastoma [10] Although extensive resection is usually recommended, permanent numbness will occur at the region innervated by the mental nerve owing to the excision of the inferior alveolar nerve so the resection of the nerve should be avoided whenever possible [11].

In regards to the fact that Nakamura et al. have found no invasion of the nerve or nerve bundle by ameloblastoma, they recommend a more conservative approach and proper follow up if the canal wall is not damaged [11]. A method of preservation of the IAN when the bony canal wall is intact has been reported by Ishikawa et al. [12]. However, Nakamura et al. found that even when the bony wall of the mandibular canal was destroyed, the ameloblastoma was clearly separated from the nerve by bone or connective tissue [11]. Wong et al. have recommended when preservation of the nerve is highly desirable; this technique maybe a procedure to be considered in certain circumstances [7].

Furthermore, clinical features may give more information about the growth characteristics of tumors involving the inferior alveolar nerve. Mental neurosensory disturbance generally indicates a possibility of damage to the inferior alveolar nerve by the tumor. Tingchun et al. suggested the resection of the nerve if the mandibular canal was involved and neurological symptoms were present [11,13]. Engelbret et al. The Involvement of the IAN was not dependent on the gross or histological type. reported that most ameloblastomas had a mixed histological growth pattern, but neural involvement occurred irrespective of the histological variant. Small ameloblastomas, well delineated from the IAN, may well be treated differently [10]. Although the size of Plexiform Ameloblastoma was not small, and also patient had a partial neurosensory disturbance, we preserved the IAN in this presented case. Patient has no any neurosensory disturbance in three months after resection.

Transport distraction osteogenesis (TDO) may be the effective treatment of option for reconstructing mandibular lateral defects after the radical resection of ameloblastomas, to complete the restoration of form and function, dental rehabilitation is also required due to the loss of dentition. Currently, the plate-guided transport distraction device (PGTD) is used as a reconstruction plate which is placed to fill the mandibular lateral defects [9]. It has some advantages over microvascular free tissue transfer or non-vascularised bone grafts. The iliac crest is the most common donor site for non-vascularised bone, but, although it has a high success rate, related donor-site morbidity can occur ( [14] Free flaps have a high success rate and are useful in the reconstruction of facial symmetry and in restoring missing tissues including large regions of bone and soft tissue defects. Microvascular free tissue transfer is technically aggressive and challenging with significant donor site morbidity. Free flaps often result in inadequate bone for mandibular rehabilitation with osseointegrated dental implants [9,15]. Because of these reasons we decided that using PGTD for reconstruction after radical resection.

Preoperative treatment planning is important to determine the correct placement and to apply the correct vector because implementing transport distraction osteogenesis around a curve of the mandible body presents specific challenges. The PGTD allows significant vector control in all plane of space by following the reconstruction plate. Although the distractor moves along the curve, the regenerated tissue could straighten wrongly as like a line. For this reason, a surgical acrylic splint can be placed before the activation of the distractor and it helps to guide the transported bone superiorly and, more importantly, to support the desired curve. Alternatively, the splint can be placed after distraction whereas the regenerated tissue can still be molded before ossification [16]. In contrary, even we did not conduct any splint intraorally for implementing correct vector, thanks to the PGTD which was desired via a stereolithographic 3-D model ensured well curve and complete ossification.

Single step or double steps of TDO can be performed by surgeons. The first step of TDO was horizontal distraction and reconstruction of the mandibular body, and the second step was vertical distraction and restoration of the height of the ramus. Although the reliable results obtained using just single (first) step TDO, to reconstruct large mandibular defects involving the body, angle, and the ramus used internal devices. For these reasons, the use of double-step TDO which performing first and second stage together can achieve effective results. In contrast, double-step TDO requires a long period for the overall treatment period and second surgery with increasing risk of morbidity for double-step TDO are limitations when it is used for reconstructing mandibular defects [17]. In a similar reason, our patients were not eager to have a second surgery and prolong treatment process. Besides she was satisfied her appearance so we did not decide to perform double-step TDO.

Although PGTD looks like a promising procedure of the reconstruction of maxillomandibular defects with great clinical results, it has the disadvantage of prolonged time of treatment, follow up, and the use of big transport devices. The future of bone transport relies on innovations that will accelerate the mineralization of new bone regenerate and biomechanical developments in the device design, may it be a minimized transport device for better patient conformity [16,18].

When the healing of the segments of the mandible is completed, osseointegrated dental implants (OI) with the dental prosthesis are recommended in order to finalize the restoration, which is currently the gold standard especially thanks to the new bone is similar to the native bone, so it is ideal for dental implant or prosthetic rehabilitation [2,19]. Titanium dental implants are essential for prosthetic rehabilitation and for maintaining the bulk of regenerated tissue and decreasing the rate of bone resorption. For this reason, the treatment methods include over-distraction and placement of osseointegrated dental implants [9].

#### 4. Conclusion

In the posterior mandible, the IAN often lies adjacent to or is entrapped within the tumor. In such cases, a decision must be made to preserve or to resect the nerve. Especially in young patients, the inferior alveolar nerve can be protected. Because our patient was young, we preferred to preserve the nerve, and no recurrences occurred during the seven-year follow-up period. In conclusion, for the further treatment options of Ameloblastomas, especially in young patients, the inferior alveolar nerve can be preserved. Therefore the reconstruction of the mandible with single stage plate-guided transport distraction osteogenesis can be used to provide bone contour simultaneously and rehabilitation of the dentition with dental implants after the treatment of ameloblastoma with resection.

## Conflicts of interest

Authors declare that they have no conflict of interest.

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