Rehabilitation of recurrent unicystic ameloblastoma using distraction osteogenesis and dental implants

M Natashekar, R Chowdhary¹, NK Chandraker²

Private Practice, Hosur Road, Bangalore, ¹Branemark Osseointegration Center India, Golden Plaza Complex, Court Road, Gulbarga, ²Department of Prosthodontics, Government Dental College, Raipur, India

Abstract

Ameloblastoma is a true neoplasm of odontogenic epithelial origin. Surgical resection of the ameloblastoma is well-documented and an accepted treatment modality. Vertical distraction of the alveolar process is an efficient method for augmentation. This method of providing additional bone and soft tissue for implant placement is becoming more common. This clinical report describes the use of distraction osteogenesis and fixed implant supported prosthesis to treat a postsurgical alveolar defect as a result of the resection of a unicystic ameloblastoma in the anterior mandibular region. As a result of alveolar distraction a segment of mature bone was transported vertically in order to lengthen the crest, for better implant anchorage. Further clinical and experimental studies of the technique with long-term follow-up are needed, to confirm bone and implant stability, as it relates to alveolar height.

Key words: Dental implant, distraction device, segmental resection, unicystic ameloblastoma

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Introduction

Ameloblastoma is a benign, slow growing, odontogenic neoplasm. It is the second most common odontogenic neoplasm and only odontoma outnumbers it in the reported frequency of occurrence.[1-3] The average age at diagnosis is consistently reported as being in the range of 33 to 39 years and most cases cluster between the age of 20 and 60 years.[1-5] It mainly affects the mandible, but varies among racial groups.[1,6] Asians seem to have fewer tumors involving the ramus than do whites or blacks, whereas, blacks have an increased frequency of tumors in the anterior mandible compared to the other two groups.[1,3]

The nature of deficiency as a result of segmental resection may present an obstacle to ideal implant positioning, by compromising the aesthetic and prosthetic needs. To overcome this, various methods have been applied. The technique of distraction osteogenesis is becoming a routine part of the surgeon’s armamentarium. Distraction osteogenesis is defined as a biological process of bone formation occurring between the surfaces of vital bone segments, which are gradually separated by incremental traction.[7,8] This clinical study describes the use of distraction osteogenesis followed by fixed implant supported prosthesis to treat post-surgical defects formed as a result of the segmental resection of a unicystic ameloblastoma in the mandibular anterior region.

Case Report

In 2002, a 56-year-old male visited our dental hospital with a swelling in the mandibular anterior region, which was diagnosed as a Unicystic Ameloblastoma. Curettage of the lesion was done as a line of treatment. After four years, the patient revisited the hospital with an intraoral swelling in the same anterior mandibular region [Figure 1]. An orthopantomograph showed a radiolucency,
Natashekar, et al.: Reosseointegration of the recurrent unicystic ameloblastoma

approximately 1 x 2 cm in size, in relation to the apical region of the mandibular incisors and left canine [Figure 2]. A biopsy was done, confirming the diagnosis of the recurrence of Follicular Unicystic Ameloblastoma. Surgical excision of the lesion was carried out, along with the mandibular incisors and the left mandibular canine, which were periodontically compromised. Along with the tumor, 6 mm of the surrounding unaffected bone was resected, to avoid further recurrence of the lesion. An extraosseous, unidirectional, alveolar distractor was placed on the buccal surface below the resected margins [Figure 3], followed by a horizontal bone cut approximately 14 mm below the resected margin, and a vertical bone cut approximately 4 mm from the roots of the adjacent teeth, with slight convergence toward each other, in an apical direction, to allow free movement of the transport segment during distraction [Figure 4]. The flaps were replaced and sutured. After a latency period of one week, the distractor was activated by a screw pitch, 1 mm per day, for 22 days. Orthopantographs were taken at one-month intervals [Figures 5 and 6]. After a consolidation period of three months the flaps were exposed and the distractor removed. The distracted site tissue was healthy, but the transported segment was marginally tilted toward the lingual direction (due to a change in the vector) [Figure 7], and was corrected by traction for a favorable dental implant placement site, and the site was stabilized with a titanium plate [Figure 8]. This was followed by the placement of two single piece endosseous implants of dimension 4.2 x 10 mm each (TRX-OP, Hitec, Israel), along with hydroxyapatite bone grafting at the osteotomy margin [Figure 9]. The flap was replaced and sutured. A recall visit was made after two weeks [Figures 10 and 11] and a definitive impression was made using polyvinyl siloxane. A cement-retained, metal-ceramic, fixed partial denture of four units was made, which was cemented using Glass ionomer cement (Type 1, GC, Malaysia) [Figure 12]. At the two-year recall, no functional or esthetic difficulties with restoration were found, and adequate bone height was maintained as per the radiographic evaluations [Figure 13].

Discussion

Three types of ameloblastoma are distinguished based on their gross appearance — the unicystic, the multicystic, and the solid type. The unicystic ameloblastoma has a fibrous connective tissue capsule, and therefore, has a much lower rate of recurrence. The solid or multicystic ameloblastoma has a tendency to be locally invasive and has a high incidence of recurrence if not adequately removed.[9] Histopathologically, six subtypes of ameloblastoma are recognized — follicular, acanthomatous, granular cell, basal cell, desmoplastic, and plexiform.[1-3,10,11]

Although often considered benign, ameloblastoma can be aggressive locally, and proliferating lesions and malignant transformation have been reported.[12-16] Not surprisingly, treatment modalities have varied considerably. These have included simple enucleation and more radical resection with reconstruction.[17-20] In terms of a comparison of recurrence rates of different surgical modalities, relatively high recurrence rates were observed in patients treated by marsupialization followed by enucleation, with bone curettage (45.5%) and enucleation with bone curettage (18.2%).[8] Recurrence rates after radical surgery and conservative treatment were 7.1 and 33.3%, respectively.[8]

Despite the extensive literature on ameloblastoma, there is still considerable disagreement with regard to the principles of treatment of this tumor. When planning the treatment of ameloblastoma, it is important to understand the growth characteristics, so as to remove the full extent of the tumor, including the surrounding tissue. Otherwise the remaining tumor cells may lead to multiple morbidities of recurrence. Muller, based on the histopathological study of an ameloblastoma, recommend that a margin of at least 1 cm of healthy bone be resected. [21] Gardener and Pecak suggest a marginal resection, with a 1.5 cm border of apparently unaffected bone, in even small solid
multicystic ameloblastomas. In rare cases when the lesion is diagnosed early, there may be sufficient bone to resect the tumor with an adequate margin and maintain continuity of the lower border. Understanding of the biological behavior of the ameloblastoma has revealed that unicystic lesions are well localized by the fibrous capsule of the cyst, with few tumor-broaching peripheral tissues, whereas, multicystic and solid lesions are characterized by aggressive infiltration into the adjacent tissue. This suggests that surgical margins are based on the assumption of tumor behavior rather than on the histopathological studies of tumor growth and invasiveness.

After tumor resection, one of the most common problems with prosthetic rehabilitation by oral implants is that of insufficient bone height. This is often a contraindication...
for implant placement and implies that the ratio of crown to implant length is too great, a factor that will probably reduce not only the useful life span of the implant for the perspective of the biomechanical function, but also the aesthetic outcome.\textsuperscript{27,28}

Multiple reconstruction and regeneration methods have been applied in order to augment the alveolar ridge. Present day treatment for alveolar ridge reconstruction includes autogenous bone grafting,\textsuperscript{29-31} guided bone regeneration (GBR),\textsuperscript{32,33} and use of alloplastic material.\textsuperscript{32,33} When using an autogenous bone graft, donor site morbidity is unavoidable and some resorption of the bone graft occurs. The GBR technique of ridge augmentation has been extensively documented and the difficulty in providing adequate space for regeneration and obtaining sufficient bone volume is a known fact.\textsuperscript{32-34} This technique is useful for limited defects of the alveolar ridge. Alloplastic materials are not suitable for implant placement.\textsuperscript{32-34}

A useful tissue engineering technique that allows the height of the alveolar ridge to be increased effectively has gained increasing acceptance, and is called alveolar distraction osteogenesis.\textsuperscript{28,35} Distraction osteogenesis is based on the principal of ‘tension – stress,’ with gradual application of tensile forces stimulating new bone formation parallel to the vector of distraction.\textsuperscript{36,37} Vector control is vital for the precision demanded in the implant site preparation.\textsuperscript{38} In 1970, Wagner used a new distraction to 1.5 mm per day with initiation of distraction at surgery.\textsuperscript{38} In 1987 De Bastaini et al., advocated callous distraction by increasing the latency period to 14 days.\textsuperscript{35} Dr. Gavriel Ilizarov pioneered distraction osteogenesis.\textsuperscript{39} Block et al., reported the first case of alveolar distraction in beagle dogs.\textsuperscript{30} Chin and Toth were the first to describe alveolar distraction in humans, in 1996, using an internal distraction device.\textsuperscript{40} Gaggle et al. and Klein et al., demonstrated a new operative technique for alveolar ridge augmentation, using a distraction implant.\textsuperscript{41,42}
Alveolar distraction devices are of the intraosseous and extraosseous type. Following an osteotomy, activation of distraction osteogenesis takes place as it relates to alveolar height and width, is needed.

9. Careful thought should be applied and tailored to individual patients and situations, based not only on good evidence, but also on experience, availability of time and resources, and compliance. For management of ameloblastoma, the growth pattern and the specific jaw in which the tumor is found are the most important factors when considering the treatment option. A combination of onlay grafting and alveolar distraction is often needed to achieve the appropriate three-dimensional reconstruction of the segmental defect of the alveolar bone. Further study of the technique, with a long-term follow-up to confirm bone and implant stability, as it relates to alveolar height and width, is needed.

**Summary**

Careful thought should be applied and tailored to individual patients and situations, based not only on good evidence, but also on experience, availability of time and resources, and compliance. For management of ameloblastoma, the growth pattern and the specific jaw in which the tumor is found are the most important factors when considering the treatment option. A combination of onlay grafting and alveolar distraction is often needed to achieve the appropriate three-dimensional reconstruction of the segmental defect of the alveolar bone. Further study of the technique, with a long-term follow-up to confirm bone and implant stability, as it relates to alveolar height and width, is needed.

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