Rehabilitation of recurrent unicystic ameloblastoma using distraction osteogenesis and dental implants

M Natashekar, R Chowdhary[1], NK Chandraker[2]

Private Practice, Hosur Road, Bangalore, [1]Branemark Osseointegration Center India, Golden Plaza Complex, Court Road, Gulbarga, [2]Department 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 post-surgical 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

Date of Acceptance: 27-Feb-2011

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.[3,6] 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 radioluscency,

Address for correspondence:
Dr. Ramesh Chowdhary,
Branemark Osseointegration Center India, Golden Plaza Complex, Court Road, Gulbarga-585 102, India.
E-mail: drramc@yahoo.com

Access this article online

Quick Response Code:

Website: www.njcponline.com

DOI: 10.4103/1119-3077.91763

PMID: 22248957

approximately 1 × 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 where 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 × 10 mm each (TRX-OP, Hitec, Isreal), along with hydroxyapitite bone grafting at the osteotomy margin [Figure 9]. The flap was replaced and sutured. A recall visit was made after two weeks [Figures10 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].

Figure 1: Intraoral swelling in relation to 31 and 32

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 marsupalization 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

Figure 2: Orthopantomography showing radiolucency in relation to 31 and 32

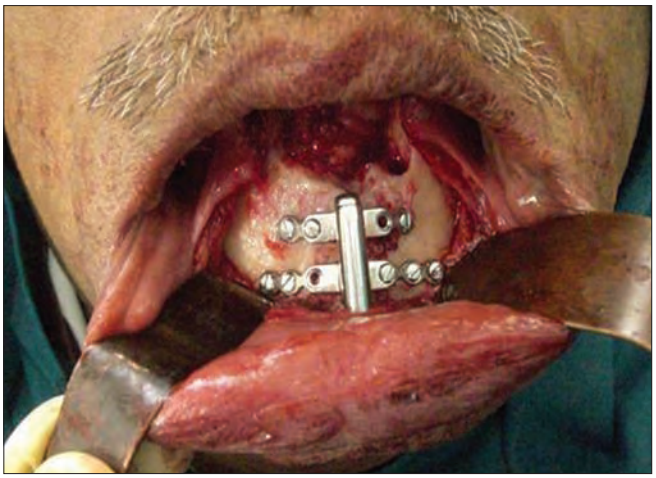

Figure 3: Positioning of the Alveolar distractor

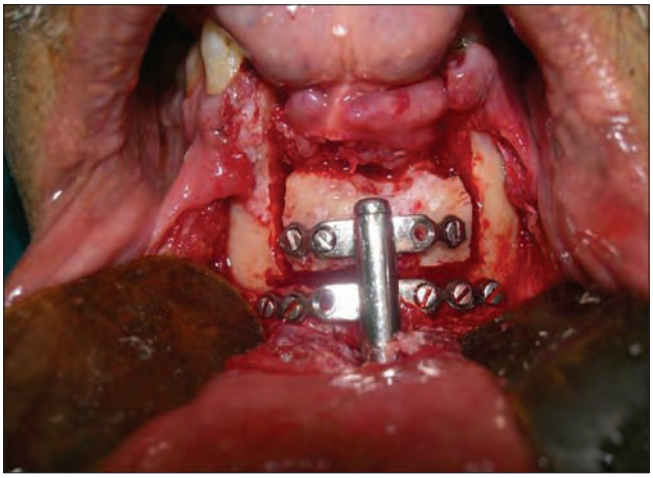

Figure 4: Vertical and horizontal sections of the bone with distractor in position

Figure 5: Radiograph showing the Alveolar distractor in position

Figure 6: Radiograph showing the vertically moved bone, due to distraction

Figure 7: Lingual tilt of the transported segment

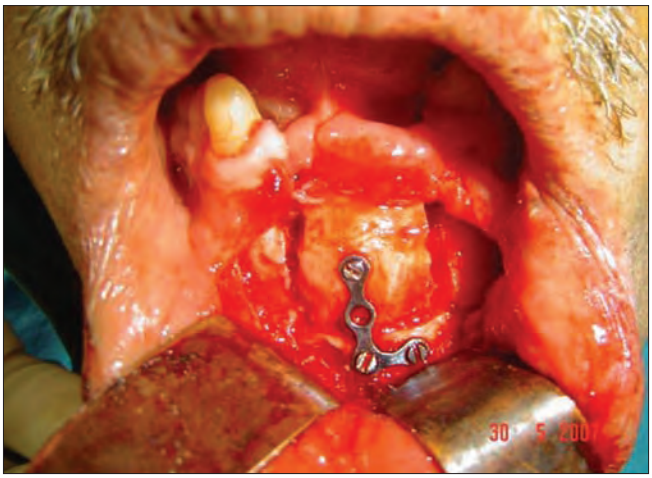

Figure 8: Correction of the lingually tilted segment by traction using a titanium plate

multicystic ameloblastomas.[22] 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.[23] 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.[24,25] This suggests that surgical margins are based on the assumption of tumor behavior rather than on the histopathological studies of tumor growth and invasiveness.[26]

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

Figure 9: Placement of Endosseous Single piece implants

Figure 11: Four unit metal ceramic fixed prostheses in position

Figure 13: Radiograph after a two-year follow-up

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.[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

Figure 10: Radiograph showing the implants placed in the transported segment of the bone

Figure 12: Radiograph showing the restored implants

autogenious bone grafting,[29-31] guided bone regeneration (GBR),[32,33] and use of alloplastic material.[32,33] When using an autogenious 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.[32-34] This technique is useful for limited defects of the alveolar ridge. Alloplastic materials are not suitable for implant placement.[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.[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.[36,37] Vector control is vital for the precision demanded in the implant site preparation.[38] In 1970, Wagner used a new distraction to 1.5 mm per day with initiation of distraction at surgery.[38] In 1987 De Bastaini *et al.*, advocated callous distraction by increasing the latency period to 14 days.[38] Dr. Gavriel Ilizarov pioneered distraction osteogenesis.[39] Block *et al.*, reported the first case of alveolar distraction in beagle dogs.[38] Chin and Toth were the first to describe alveolar distraction in humans, in 1996, using an internal distraction device.[40] Gaggle *et al.* and Klien *et al.*, demonstrated a new operative technique for alveolar ridge augmentation, using a distraction implant.[41,42]

Alveolar distraction devices are of the intraosseous and extraosseous type.[38] Following an osteotomy, activation of a distractor device leads to the formation of a gap between segments. Furthermore, a regenerate formed between the bones has four zones from the center to the periphery of — fibrous tissue, extended bone formation, bone remodeling, and mature bone.[44,47] Distraction osteogenesis takes place in four clinical phases — latency, distraction, stabilization, and distraction removal.[38] A latency period of two to five days is indicated in young patients and adults, when minimal surgical trauma is encountered. A period of seven to fourteen days is recommended in older patients or when increased surgical trauma is noted.[48,51]

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.

References

1. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. Eur J Cancer B Oral Oncol 1995;31:86-99.
2. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and maxillofcail pathology. 2nd ed. Philadelphia: WB Saunders; 2002. p. 611-9.
3. Kessler HP, Schwartz-Dabney C, Ellis E 3rd. Recurrent left mandibular enlargement. J Contemp Dent Pract 2003;3:127-37.
4. Zawhlen RA, Gratz KW. Maxillary ameloblastomas: A review of the literature and of a 15 year database. J Cranio Maxillofac Surg 2002;30:273-9.
5. Henderson JM, Sonnet JR, Schlesinger C, Ord RA. Pulmonary metastasis of ameloblastoma: Case report and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;88:170-6.
6. Raghoebar GM, Liem RS, Vissink A. Vertical distraction of the severely resorbed edentulous mandible: A clinical, histological and electron microscopic study of 10 treated cases. Clin Oral Implant Res 2002;13:558-65.
7. Ilizarov GA. Clinical application of the tension-stress effect for limb lengthening. Clin Orthop Relat Res 1990;250:8-26.
8. Batal SM, Cottrell DA. Alveolar distraction osteogenesis for implant site development. Oral Maxillofac Surg Clin N Am 2004;16:91-109.
9. Williams TH. Management of ameloblastoma: A changing perspective. J Oral Maxillofac Surg 1993;51:1064-70.
10. Tsaknis PJ, Nelson JF. The maxillary ameloblastoma: An analysis of 24 cases. J Oral Surg 1980;38:336-42.
11. Keszler A, Paparella ML, Dominguez FV. Desmoplastic and non-desmoplastic ameloblastoma: A comparative clinicopathologicla analysis. Oral Dis 1996;2:228-31.
12. Small IA, Waldron CA. Ameloblastomas of the jaws. J Oral Surg 1955;8:281-97.
13. Phillips SD, Corio RL, Brem H, Mattox D. Ameloblastoma of the mandible with intracranial metastasis to the lungs and lymph nodes: A case study. Arch Otolaryngol Head Neck Surg 1992;118:861-3.
14. Ueda M, Kaneda T, Imaizumi M, Abe T. Mandibular ameloblastoma with metastasis to the lungs and lymph nodes: A case report and review of the literature. J Oral Maxillofac Surg 1989;47:623-8.
15. Corio RL, Goldblatt L, Edwards P, Hartman K. Ameloblastic carcinoma: A clinicopathologic study and assessment of eight cases. Oral Surg Oral Med Oral Pathol 1987;64:570-6.
16. Slootweg PJ, Muller H. Malignant ameloblastoma or ameloblastic carcinoma. J Oral Surg 1984;57:168-76.
17. Feinberg SE, Steinberg B. Surgical management of ameloblastoma: Current status of the literature. Oral Surg Oral Med Oral Pathel Oral Radiol Endod 1996;81:383-8.
18. Muller H, Slootweg PJ. The ameloblastoma: The controversial approach to therapy. J Maxillofac Surg 1985;13:79-84.
19. Williams TP. Management of ameloblastoma: A changing perspective. J Oral Maxillofac Surg 1993;51:1064-70.
20. MacIntosh RB. Aggressive surgical management of ameloblastoma. Oral Maxillfac Surg Clin North Am 1991;3:73-97.
21. Muller, H. Het ameloblastoom. Een klinisch en histopathologisch onderzoek. Thesis. Utrecht, The Netherlands: 1983.
22. Gardner DG, Pecak AM. The treatment of ameloblastoma based on pathologic and anatomic principles. Cancer 1980;46:251-9.
23. Ord RA, Blanchaert RH Jr, Nikitakis NG, Sauk JJ. Ameloblastoma in children. J Maxillofac Surg 2002;60:762-70.
24. Gardner DG. A pathologist's approach to the treatment of ameloblastoma. J Oral Maxillofac Surg 1984;42:161-6.
25. Nakamura N. Clinical and histopathological studies on the characteristics of growth of mandibular ameloblastoma. Jpn J Oral Maxillofac Surg 1991; 37:1600-15s.
26. Gortzak RA, Latief BS, Lekkas C, Slootweg PJ. Growth characteristics of large mandibular ameloblastomas: Report of 5 cases with implication for the approach to surgery. Int J Oral Maxillofac Surg 2006;35:691-5.
27. Garcia-Garcia A, Somoza-Martin M, Gandara-Vila P, Saulacic N, Gandara-Rev JM. Alveolar distraction before insertion of dental implants in the posterior mandible. Br J Oral Maxillofac Surg 2003;41:376-9.
28. Mazzonetto R, Allais De Maurette M. Radiographic evaluation of alveolar distraction osteogenesis: Analysis of 60 cases. J Oral Maxillofac Surg 2005;63:170-1.
29. Nystrom E, Kahneberg KE, Gunne J. Bone grafts and Branemark implants in the treatment of severely resorbed maxilla: A two – year longitudinal study. Int J Oral Maxillofac Impl 1993;8:45-53.
30. Satow S, Slagter AP, Stoelinga PJ, Habets LL. Interposed bone grafts to accommodate endosteal implants for retaining mandibular over dentures: A 1-7 year follow-up study. Int J Oral Maxillofac Surg 1997;26:358-64.
31. Triplett RG, Schow SR. Autologous bone grafts and endosseous implants: Complementary techniques. J Oral Maxillofac Surg 1996;54:489-94.
32. Caplanis N, Sigurdsson TJ, Rohrer MD, Wikesjö UM. Effect of allogeneic, freeze-dried, demineralized bone matrix on guided bone regeneration in supraalveolar peri-implant defects in dogs. Int J Oral Maxillofac Impl 1997;12:634-42.
33. Jensen OT, Greer RO Jr, Johnson L, Kassebaum D. Vertical guided bone-graft augmentation in a new canine mandibular model. Int Oral Maxillofac Implants 1995;10:335-44.
34. Rachmiel A, Srouji MP. Alveolar ridge augmentation by distraction osteogenesis. Int J Oral Maxillofac Surg 2001;30:510-7.
35. Jensen OT, Cockrell R, Kuhike L, Reed C. Anterior maxillary alveolar distraction soteogenesis: A prospective 5- year clinical study. Int J Oral Maxillofac Implants 2002;17:52-68.
36. Iliazarov GA. The tension stress effect on the genesis and growth of tissues: Part I: The influence of stability of fixation and soft tissue preservation. Clin Orthop Relat Res 1989;238:249-81.
37. Iliazarov GA. The tension stress effect on the genesis and growth of tissues. Part II. The influence of the rate and frequency of distraction. Clin Orthop Relat Res 1989;239:263-85.
38. Batal HS, Cottrell DA. Alveolar ditraction osteogenesis for implant site development. Oral Maxillofac Surg Clin North Am 2004;16:91-109.
39. Ilizarov GA. The principles of the Iliazrov method. Bull Hosp Joint Dis 1988;48:11.
40. Chi M, Toh BA. Distraction osteogenesis in maxillofacial surgery using internal devices: Report of five cases. J Oral Maxillofacial Surg 1996;54:45-53.
41. Gaggl A, Shultes G, Karcher H. Distraction implants: A new operative technique for alveolar ridge augmentation. J Craniomaxillofac Sug 1999;27:214-21.
42. Gaggi A, Shultes G, Karcher H. Distraction implants-a new possibility for

augmentative treatment of the edentulous atrophic mandible: Case report. Br J Oral Maxillofac Surg 1999;37:481-5.
43. Karp NS, McCarthy JG, Schreiber JS, Sissons HA, Thorne CH. Membranous bone lengthening: A serial histological study. Ann Plast Surg 1992;29:2-7.
44. Hartman KS. Granular-cell ameloblastoma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1974;38:241-53.
45. Chin M. Distraction osteogenesis of dental implants. Atlas Oral Maxillofac Surg Clin North Am 1999;7:41-63.
46. Klesper B, Lazar F, Siessegger M, Hidding J, Zoller JE. Vertical distraction osteogenesis of fibula transplants for mandibular reconstruction a preliminary study. J Craniomaxillofac Surg 2002;30:280-5.
47. Lucas RB. Pathology of Tumours oft4he Oral Tissues. 4th ed. London: Churchill Livingston; 1984. p. 31-60.
48. Laurie SW, Kaban LB, Mulliken JB. Donar site morbidity after harvesting rib and illic bone. Plast Reconstr Surg 1984;73:933-8.
49. Jensen OT, Cockrell R, Kuhike L, Reed C. Anterior maxillary alveolar distraction osteogensis: A prospective 5 years clinical study. Int J Oral Maxillofacial Implants 2002;17:52-81.
50. Wagner H. Operative beinverlangerung. Chirurgie 1971;42:260-6.
51. De Bastiani G, Aldegheri R, Renzi-Brivio L, Trivella G. Limb lengthening by callus distraction (callotasis). J Pediatr Orthop 1987;7:129-34.

How to cite this article: Natashekar M, Chowdhary R, Chandraker NK. Rehabilitation of recurrent unicystic ameloblastoma using distraction osteogenesis and dental implants. Niger J Clin Pract 2011;14:486-91.
Source of Support: Nil, **Conflict of Interest:** None declared.