New Regenerative Surgical Treatment of Cystic Diseases of the Jaw by Utilizing Grafting of Cancellous Iliac Bone and Replanting of Patient's Teeth

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The authors developed a new regenerative surgical technique for cystic diseases of the jaw. In this surgery, all teeth that are rooted in or located adjacent to the cyst are extracted for replanting, and attached soft tissues, including cystic wall as well as dental pulp, are completely removed. Gingiva is cut open at the alveolar crest and abraded subperiosteally, the jaw on the cystic lesion is widely exposed, and cortical bone on the frontal wall of the cystic lesion is dissected. After the entire cyst is exposed, it is completely removed with scrapers and bars. The defect is reconstructed by the following procedures: the extracted and treated teeth are replanted; cancellous iliac bone (bone marrow) is grafted around the teeth and in the defect; and after thorough washing and making numerous small holes, the frontal wall of the cortical bone is returned to the original position and fixed. Three to 4 months later when the jaw has regenerated and the replanted teeth have taken, a prosthesis is set on the replanted teeth and biting is started. This technique was applied to a 16-year-old male patient who had odontogenic keratocysts on the lower right teeth (numbers 6, 5, 4, 3, 2, and 1), the lower left teeth (numbers 1, 2, 3, and 4), and the upper right teeth (numbers 5, 4, 3, and 2). The patient regained normal biting capability, and the esthetic outcome was also satisfactory. This technique would be a useful treatment method for cystic diseases of the jaw.

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Cystic diseases of the jaw, particularly odontogenic keratocysts and ameloblastoma, that have a high frequency of recurrence are usually treated with cystectomy and fenestration[1,2] or jaw transection.[3] The resultant defect of the jaw, including loss of teeth and alveolar bone, becomes large based on the size of the cyst, and this significantly impairs the biting capability of the patient. Generally, these patients receive a denture and/or bridge as a means of mastication, but sufficient recovery is hard to obtain if the denture and bridge do not have stable support because of a large defect of the jaw. In addition, the patients are usually quite young, and those surgical treatments could significantly lessen their quality of life not only in terms of mastication but in terms of esthetic, mental, and psychological aspects.

The authors have succeeded in regenerative treatments using cancellous iliac bone (bone marrow) in the treatment of mandibular bone defects after surgical resection of oral malignant tumor and in the treatment of alveolar bone defects caused by severe periodontosis.[4,5] The authors applied this concept to the treatment of cystic diseases of the jaw and developed a new regenerative surgical technique.

Methods

Surgical Procedures

As the first step, teeth that are rooted in or located adjacent to the cyst are extracted. Soft tissues, including the cystic wall, are completely removed from the teeth, and dental pulp is also totally removed. Through an incision made on the alveolar crest, gingiva and membrane on the cheek side are subperi-

osteally dissected and the jaw on the cystic lesion is widely exposed. The site of the cyst is marked on the jaw by referring to radiographs and computed tomography images. Cortical bone on the frontal wall of the marked area is dissected using a bar and/or bone saw, and the entire cyst is exposed (Fig 1).

The second step is total resection of the cyst. Cystic tissues are completely removed from the bone surface using scrapers, and the cystic wall and bone surface that appeared after teeth extraction are also thoroughly removed using bars. This total removal is also performed on the dissected frontal wall of the jaw. After cystectomy, the defect area and the dissected frontal wall of the jaw are carefully washed several times with physiological saline solution to wash out the cystic tissues completely.

In the third step, the teeth that were extracted during the first step are replanted in the original position (the teeth could be planted in another place). The required volume of cancellous iliac bone is collected from the patient and used to fill the space around the teeth. On the dissected frontal wall of the jaw, numerous small holes are made to ensure the regeneration of blood circulation to the grafted cancellous bone. The treated frontal wall of the jaw is then returned to the original position and fixed with a wire (see Fig 1). The surgery is completed by suturing the gingiva.

After 3 or 4 months when the replanted teeth and grafted cancellous iliac bone have completely taken, a prosthesis is set on the replanted teeth and normal biting is started.

Results

A 16-year-old male patient who had odontogenic keratocysts on the lower right teeth (numbers 6, 5, 4, 3, 2, and 1), lower left teeth (numbers 1, 2, 3, and 4), and upper right teeth (numbers 5, 4, 3, and 2) (Fig 2A) received surgical treatment using our technique. The patient previously received drainage at another clinic, and pus had drained out from that point. Our surgical procedures were fully explained to the patient and his parents, and their informed consent was obtained.

On the mandible, the lower right teeth (numbers 6, 5, 4, and 3) were extracted for replanting, the cortical bone on the frontal wall of the cystic lesion was dissected, and the cyst was removed. The root of each of the lower right teeth (numbers 2 and 1) and the lower left teeth (numbers 1, 2, 3, and 4) seemed radiographically and macroscopically only slightly or almost touched by the cyst. Therefore, those teeth remained unextracted, and only the apical area of each tooth was resected (see Fig 2B). At cystectomy, the right mandibular nerve was exposed when the bone surface was removed. Because of this, careful

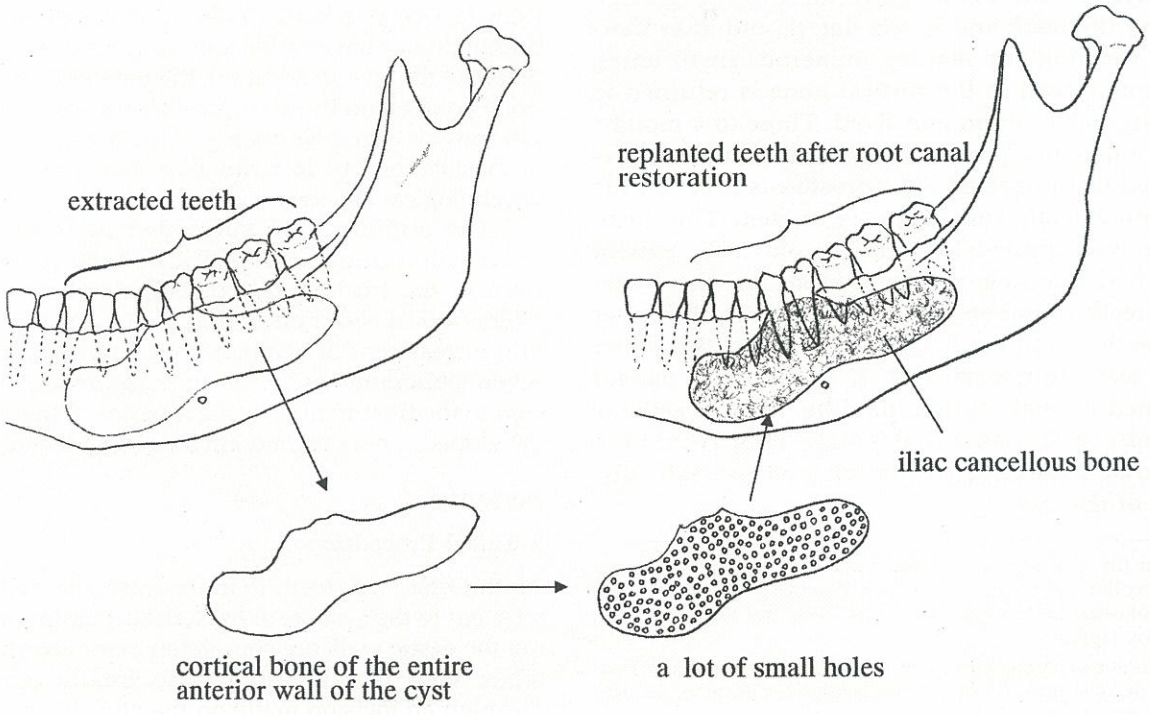
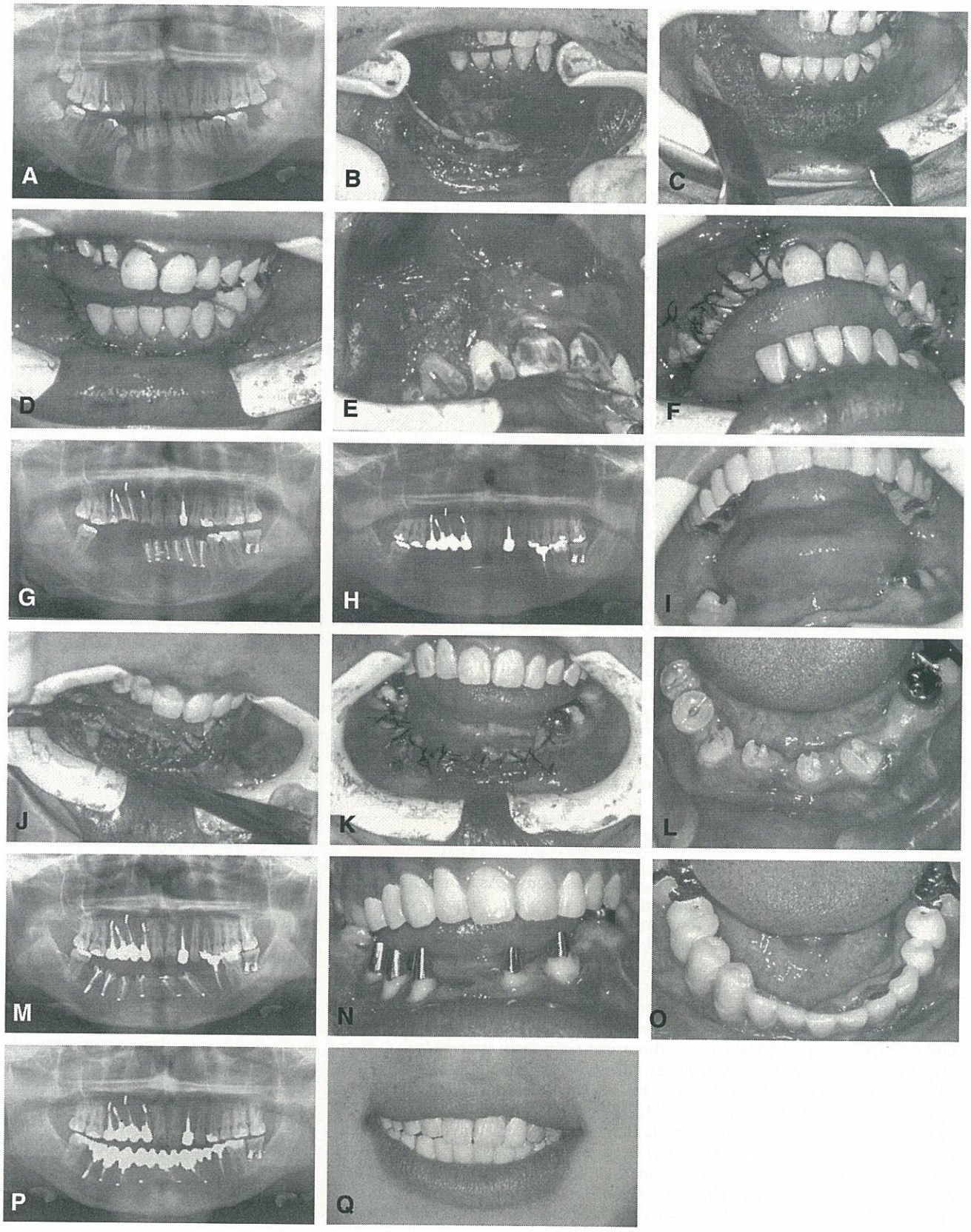


Fig 1 A diagram showing surgical procedures.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monitoring of recurrence was thought to be necessary, and the lower right teeth (numbers 6, 5, 4, and 3) were not replanted at this initial surgery. These four teeth were immersed in 30% glycerin preservative and cryopreserved at −135°C for a second operation. Cancellous iliac bone (bone marrow) was used to fill in the entire space after cystectomy (see Fig 2C), the dissected frontal wall was returned to the original position, and the gingiva was sutured closed (see Fig 2D).

On the maxilla, the right teeth (numbers 5, 4, 3, and 2) were extracted, the cyst was completely removed, and the teeth were replanted together with grafting of cancellous iliac bone (see Fig 2E-G).

After the initial surgery, the maxilla cured smoothly without any problems, but on the mandible, the right teeth (numbers 2 and 1) and the left teeth (numbers 1, 2, 3, and 4) developed pus drainage 3 weeks after the surgery. Because those teeth underwent only apical removal, infection caused by remaining cystic tissues was strongly suggested. As a treatment, those teeth were extracted; debridement, including the alveolar bone and grafted bone, was performed; and the area was made into an open wound. From the extracted teeth, soft tissue attachments and dental pulp were completely removed, and these teeth and the other four teeth were cryopreserved.

The open wound healed in about 3 weeks, and the subsequent course was carefully monitored for about 6 months; during this time, the scar tissue became softer. There was no recurrence on the site of the exposed right mandibular nerve or on the lower right teeth (numbers 2 and 1) or lower left teeth (numbers 1, 2, 3, and 4) (see Fig 2H, I). After this 6-month period, the six cryopreserved teeth were replanted in the position of the lower right teeth (numbers 6, 5, 4, 3, 2, and 1) and the lower left teeth (numbers 1, 2, 3, and 4), and the cancellous iliac bone that was collected from the patient's other side was grafted around the root of the replanted teeth (see Fig 2J, K).

The patient has had a good postoperative course, and all six teeth that were replanted during the secondary surgery have taken well (see Fig 2L, M). Four months later, a prosthesis was set. At that time, lower right tooth number 1 was fallen and a bridge was prepared to fill that space (see Fig 2N, O).

To date, 1 year and 5 months has passed since the cystectomy, 8 months since the secondary replanting of the teeth, and 3 months since setting the prosthesis and bridge. Normal eating has been regained, and the esthetic result is also quite satisfactory (see Fig 2P, Q).

Discussion

Our surgical technique aims at reproducing almost the same jaw structure and obtaining sufficient biting capability, and this is different from conventional treatments for cystic diseases[1–3] such as odontogenic keratocysts and ameloblastoma. It is well known that cancellous iliac bone contains numerous components of bone marrow and possesses the capability of bone regeneration.[6,7] In the field of maxillofacial surgery, cancellous iliac bone is commonly used as a graft to the cleft of the maxilla in patients with cleft lip and palate.[8,9] The authors have conducted regenerative surgical treatment by grafting cancellous iliac bone and have achieved good clinical and esthetic outcomes in patients with a large defect on the mandible after malignant tumor resection or with a large defect

Fig 2 A 16-year-old male patient with odontogenic keratocysts. (A) Preoperative radiograph. Odontogenic keratocysts were depicted on the lower right teeth (numbers 6, 5, 4, 3, 2, and 1), lower left teeth (numbers 1, 2, 3, and 4), and upper right teeth (numbers 5, 4, 3, and 2). (B) After cystectomy. As a preparation, the upper right teeth (numbers 6, 5, 4, and 3) were extracted for replanting and the cortical bone on the frontal wall of the jaw on the cystic lesion was dissected. (C) Cancellous iliac bone (bone marrow) was used to fill in the space after cystectomy. (D) Mandibular gingiva was sutured closed. (E) On the positions of the upper right teeth (numbers 5, 4, 3, and 2), the extracted teeth were replanted and cancellous iliac bone was grafted after cystectomy. (F) Maxillary gingiva was sutured, and the replanted teeth were fixed. (G) Radiograph 1 week after the surgery. (H) Radiograph 4 months after the surgery. Infection occurred on the root of the lower right teeth (numbers 2 and 1) and the lower left teeth (numbers 1, 2, 3, and 4). Those teeth were extracted for replanting, and debridement was performed. (I) Six months after the debridement. Infection on the mandible was healed, and a prosthesis was already set on the upper right teeth (numbers 5, 4, 3, and 2). (J) At the second operation 6 months after the initial surgery, six teeth were replanted to the mandible and cancellous iliac bone (bone marrow) was grafted. (K) Mandibular gingiva was sutured, and the replanted teeth were fixed. (L) Mandibular gingiva 4 months after the second operation. The six teeth have taken. (M) Radiograph 4 months after the second operation. (N) Preparation for setting the prosthesis. During this treatment, the replanted tooth on the lower right (number 1) fell. The other teeth on the lower right (numbers 5, 4, 3, and 2) were stable. (O) The prosthesis was set, and a bridge was prepared to fill the space of the lost tooth. (P) Radiograph 1 year and 3 months after the initial surgery and 7 months after the second surgery. Bone regeneration was confirmed, and the jaw had regained almost the same shape as before surgery. (Q) One year and 3 months after the initial surgery. The patient was satisfied with the functional and esthetic outcomes.

of alveolar bone caused by severe periodontal disease.[4,5] This treatment concept was applied to treatments for cystic diseases.

The conventional treatments for cystic diseases such as odontogenic keratocysts and ameloblastoma are fenestration and cystectomy. The biggest concern regarding these treatments is recurrence as a result of remaining cystic tissues.[10,11] Because of this, some physicians recommend radical surgical resection such as jaw transection.[3] This surgery is associated with significant physical invasion, however, and requires bone graft with a vascular pedicle for mandibular reconstruction.

To prevent recurrence, the authors conducted three procedures: 1) the cortical bone on the frontal wall of the jaw at the cystic lesion was dissected to produce a wide field for cystectomy, which remained an unblinded area and facilitated complete resection of the cyst; 2) through this wide field, the entire bone surface that touched the cyst was removed using bars, and this facilitated removal of invisible cystic tissues, and 3) after cystectomy, the open space and the dissected cortical bone were washed several times with physiological saline solution, and all cystic tissues were completely removed. These three treatments prevented recurrence of benign cystic disease. If there is concern about future recurrence because nerves are maintained, the extracted teeth from that location could be cryopreserved until the possibility of recurrence is totally removed, and the teeth could be replanted during a second operation. In our patient, postoperative infection that would be attributable to remaining cystic tissue occurred on the lower six teeth that were not extracted at the initial surgery. Because of this experience, all teeth that touch the cyst even slightly should be extracted, treated, and replanted in a future operation.

Another problem associated with conventional treatments is the loss of normal biting capability in the area of the cystectomy. Patients usually wear a denture or bridge; however, it becomes unstable when a wide area is resected, and this produces distortion of biting capability. Our patient would not regain normal biting capability if any of the conventional treatments were performed on the mandible (right teeth numbers 6, 5, 4, 3, 2, and 1 and left teeth numbers 1, 2, 3, 4) even if a denture, bridge, and/or implant were applied. In addition, patients are usually teenagers. Therefore, not only biting capability but esthetic outcome has a significant influence on their mental and psychological well-being. With our technique, alveolar bone is regenerated through the grafting of cancellous iliac bone, and almost normal biting capability is regained through replanting of the patient's teeth. These advantages provide new solutions to the above-mentioned concerns.

Another point to discuss is the loss of replanted teeth because of root resorption.[12,13] The authors have replanted 155 teeth in 30 patients with severe periodontal disease and have monitored these patients for 5 to 6 years after surgery. In their experience, there have been no patients whose replanted teeth dropped because of root absorption.[5] The authors have not yet clarified the factors that contribute to this good outcome, but one possibility is that the large volume of grafted bone marrow around the replanted teeth prevents root absorption. The authors also consider that implants on the site of replanted teeth would be possible if the teeth were lost.

With our technique, patients with a wide cystic lesion could regain normal biting capability with minimum surgical invasion. To date, there is only one patient who has been treated using our technique, but this case presents sufficient evidence that our technique would change the common understanding of surgical treatments for cystic diseases and benign tumors of the jaw.

References

1. Fedtofte P, Praetorius F. Recurrence of the odontogenic keratocyst in relation to clinical and histological features. A 20-year follow up study of 72 patients. Int J Oral Surg 1979;8:412–420
2. Browne RM. The odontogenic keratocyst. Clinical aspects. Br Dent J 1970;128:225–231
3. Ahlfors E, Larsson A, Sjogren S, et al. The odontogenic keratocyst: a benign cystic tumor? J Oral Maxillofac Surg 1984;42:10–19
4. Kiyokawa K, Tai Y, Tanaka S, et al. A new regenerative approach to oromandibular reconstruction after the resection of head and neck malignant tumor. J Craniofac Surg 2002;13:337–346
5. Kiyokawa K, Kiyokawa M, Hariya Y, et al. Regenerative treatment of serious periodontosis with grafting of cancellous iliac bone and gingival flaps and replanting of patients' teeth. J Craniofac Surg 2002;13:375–381
6. Taguchi Y, Pereira BP, Kour AK, et al. Autoclaved autograft bone combined with vascularized bone and bone marrow. Clin Orthop 1995;320:220–230
7. Zellin G, Alberius P, Linde A. Autoclaved bone for craniofacial reconstruction: effects of supplementation with bone marrow or recombinant human fibroblast growth factor-2. Plast Reconstr Surg 1998;102:792–800
8. Boyne PJ, Sands NR. Secondary bone grafting of residual alveolar and palate clefts. J Oral Surg 1972;30:87–92
9. Boyne PJ, Sands NR. Combined orthodontic-surgical management of residual palate-alveolar cleft defects. Am J Orthod 1976;70:20–37
10. Brannon RB. The odontogenic keratocyst. A clinicopathologic study of 312 cases. Part I. Clinical features. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1976;42:54–72
11. Pindborg JJ, Hansen J. Studies on odontogenic cyst epithelium. 2. Clinical and roentgenologic aspects of odontogenic keratocysts. Acta Pathol Microbiol 1963;58:283–294
12. Burnette WE. Fate of an iliac crest graft. J Periodontol 1972;43: 88–90
13. Dragoo MR, Sullivan HC. A clinical and histological evaluation of autogenous iliac bone grafts in humans. I. Wound healing after 2 to 8 months. J Periodontol 1973;44:599–613