ACP
AMERICAN COLLEGE OF PROSTHODONTISTS

Placement and Restoration of Dental Implants in a Patient with Paget's Disease in Remission: Literature Review and Clinical Report

Jonathan M. Rasmussen, DDS[1] & Matthew L. Hopfensperger, DDS, MS[2]

[1] Prosthodontic Resident, Mayo Clinic, Rochester, MN
[2] Director of the Pre-doctoral Dental Implant Program, Department of Prosthodontics, University of North Carolina School of Dentistry, Chapel Hill, NC

Keywords
Dental implants; low bone density; osteitis deformans; osseointegration; osseointegrated implants; Paget's disease; poor bone quality; Straumann; undersized osteotomy; Wide Neck Implants.

Correspondence
Jonathan Rasmussen, 200 First St. SW, Mayo W-4, Rochester, MN 55905. E-mail: rasmussen.jonathan@mayo.edu

This clinical report was presented at the ITI US Congress on October 24, 2004 in Chicago, IL.

Accepted April 19, 2006.

doi: 10.1111/j.1532-849X.2007.00240.x

Abstract

Paget's disease is a disorder of bone that results in the replacement of normal bone with highly vascularized, low density bone. The diminished bone quality associated with Paget's disease is a relative contraindication to the use of dental implants, as it interferes with achievement of osseointegration. There is no current literature on the success of dental implants in patients with Paget's disease. Furthermore, there is no current literature on dental implant success in patients with remissive Paget's disease or in bone that appears to be unaffected by the disease in patients with Paget's. This case report follows the treatment of a patient with a partially edentulous maxilla and mandible (ACP PDI Classification III) who presented with remissive Paget's disease. Definitive treatment included the placement of four endosseous implants used to retain full veneer crowns. Despite the radiographic appearance of adequate bone quality, upon surgical placement it was discovered that the quality of bone was poor, yet implant survival was achieved through careful surgical management and rigid splinting of some of the endosseous implants.

Osteitis deformans, or Paget's disease of Bone (PDB), is a chronic disorder of the adult skeleton in which localized areas of bone become hyperactive, resulting in replacement of the normal bony matrix with a highly vascular, softened, enlarged bone.[1] PDB is a localized bone disease that may have widespread distribution, as opposed to a generalized disease such as hyperthyroidism.[1]

Under normal physiologic conditions, the skeleton is remodeled to maintain its structural integrity. When the rate of bone turnover is increased, as in PDB, the new bone is formed with less structural order and appears on histologic examination as a disorganized mosaic of woven and lamellar bone.[2] Although bone production is disorganized and there is very rapid deposition of new bone in PDB, the primary cellular abnormality in patients with PDB resides in the osteoclasts. The osteoblasts appear to be normal but have increased activity in response to the markedly increased bone resorption.[3] The number of osteoclasts in pagetic bone can be increased by up to ten-fold, compared with normal bone. The osteoclasts of pagetic bone are also much larger than normal and may contain as many as 100 nuclei in a single cell, compared with three to ten nuclei in a normal osteoclast.[4]

The epidemiology of PDB shows a slight male predominance (male:female ratio of 3:2). It is believed to affect 2–3% of the population over the age of 50 years.[5] The disease demonstrates increasing prevalence with age.[6,7] The etiology of PDB is unclear. When first described by Paget, it was thought to be inflammatory and to have an infectious origin.[8] Current theories have focused on genetic and viral factors. The genetic theories are supported by epidemiological studies;[9,10] viral theories stem from ultrasonic studies demonstrating nuclear and cytoplasmic inclusions.[11,12] More recent studies demonstrate that the inclusions resemble paramyxoviruses.[13,14]

PDB is asymptomatic and without clinical findings in approximately 80 to 90% of those with the condition.[15,16] Among those with symptoms, the major complaint is bone pain; signs of fracture and bone deformation are also noted.[17] The jaws are affected in approximately 15% of cases. Common dental complications include malocclusion, tooth mobility, root resorption, hypercementosis, excessive bleeding on extraction,

osteomyelitis, and poorly fitting dentures.[18] Incidence is more frequent in the maxilla by a 2:1 ratio.[19]

The diagnosis of PDB is established through clinical and radiographic findings together with biochemical analysis.[19] Serum alkaline phosphatase is a biochemical marker of bone formation, and in PDB is an accurate indicator of bone turnover and disease activity.[2] The radiographic appearance of PDB depends on the stage of the disease. The resorptive phase is characterized by radiolucent lesions (ground glass appearance), and the appositional phase by irregular radiopacity (cotton-wool appearance).[19] The agents of choice for treating PDB are the bisphosphonates.[20]

Modern dental implants are placed into bone with the goal of becoming rigidly fixed to the bone in a process of osseointegration.[21–33] When bone density is low, the likelihood of achieving osseointegration diminishes.[33] The low bone density associated with PBD may therefore be considered as a relative contraindication to implant placement. No clinical reports are found in a MEDLINE search of the dental literature when using the terms "osseointegration" and "Paget's disease," although Roberts et al[34] describe it as a potential risk factor. One clinical report of auricular implants in a patient with PBD was, however, found in the otorhinolaryngologic literature.[35] The following is a clinical report on the treatment of a patient with remissive PDB using Straumann Wide Neck dental implants (WNI) (Straumann, Basel, Switzerland).

Clinical report

Background

A 71-year-old Caucasian male presented to the University of North Carolina School of Dentistry student clinics with a chief complaint of missing teeth. The patient was partially edentulous in the maxilla and mandible with a Kennedy Class II edentulous space in the maxillary arch and Kennedy Class II Modification I edentulous space in the mandibular arch (ACP PDI Class III). The patient had used a removable partial denture for a period of 6 months, but found that it was not comfortable despite numerous denture adjustments.

Treatment options included restoration with a shortened dental arch or intervention designed to address the underlying foundation through the use of implant-supported prostheses. After discussion of the risks and benefits of both treatment alternatives the patient elected to proceed with implant-supported prostheses.

Diagnostic procedures

A thorough review of the patient's medical history revealed that the patient had been diagnosed with Paget's disease approximately 12 years earlier. The patient reported that the disease was in clinical remission. Upon further investigation with the patient's physician, the patient's diagnosis of Paget's disease of the femur was confirmed.

During clinical examination it was noted that the patient was missing the third molar in the right maxilla and all molars in the left maxilla. In the mandibular arch, the patient was missing the left first and third molars while all molars were missing on the right side (Figs 1–3). The patient's dentition showed abrasive wear that was thought to be secondary to a self-reported history of clenching and bruxism. Periodontally, the patient presented with healthy soft tissues and isolated pocketing of less than 5 mm.

During the treatment planning stage, a full mouth series of periapical radiographs and a panoramic radiograph (Fig 4) were taken, and diagnostic casts were made and mounted on a semi-adjustable articulator. Radiologic consultation demonstrated that the bone in the proposed implant sites showed no signs of being affected by PDB. Treatment options were discussed and a plan was formulated. The patient was informed of the specific risks associated with dental implants and the potential for unforeseen complications associated with his Paget's disease. The patient understood the risks and possible complications and consented to restoration with dental implants.

The treatment plan consisted of restoring the maxillary left first and second molars, the mandibular left first molar and mandibular right first molar with Straumann Wide Neck implants. Based on the bone morphology, 10-mm implants were selected for the two maxillary sites, and 12-mm implants were planned for the two mandibular sites. It was decided to restore the implants with full veneer gold crowns and to splint the maxillary left first and second molars due to the patient's history of clenching and bruxing. The left mandibular second molar was not replaced, because there was insufficient interarch space for a restoration in this site.

Treatment sequence

Implant placement was accomplished using regional anesthesia — 2% Xylocaine with 1/100,000 epinephrine (AstraZeneca, Wilmington, DE). Full thickness flaps were elevated in all areas of planned implant placement except in the mandibular left molar site, where a tissue punch was used. A surgical template was used to ensure favorable implant location. Upon preparation of the osteotomies, Type IV bone (Lekholm and Zarb classification, 1985) was noted in all four sites. The implant sites for the maxillary left first and second molars as well as the mandibular right first molar were prepared using a modified surgical technique. The mandibular left first molar osteotomy site was prepared as directed by the implant manufacturer (Figs 5-7). Due to the poor bone quality, intra-operative and postoperative measures were taken to increase the chance of successful implant osseointegration. These measures included omitting the final drill in the manufacturer's suggested drilling sequence in three of the four sites (modified surgical technique), prescribing a soft diet, and waiting 6 months before loading the implants.

Implant stability was achieved in the maxillary left first and second molar sites and the mandibular right first molar site. The implant in the mandibular left first molar site, however, was determined to lack primary stability. This is the one site in which the complete recommended drilling sequence was performed, therefore resulting in a wider diameter osteotomy site than the other three implant locations. The patient was informed of the possibility of implant failure in this site and given specific postoperative instructions to improve chances of osseointegration. One week after placement, the implants were

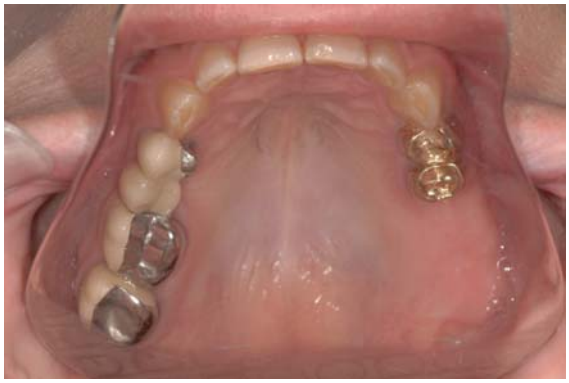

Figure 1 Preoperative maxillary occlusal view.

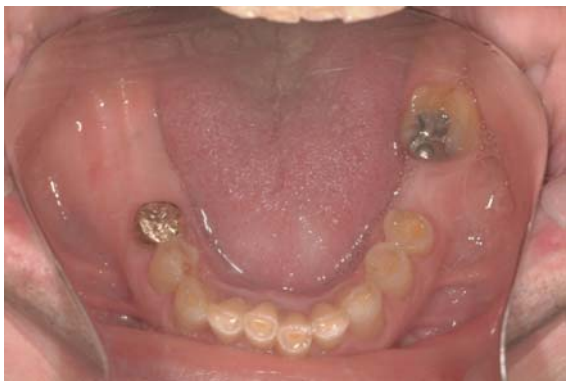

Figure 2 Preoperative mandibular occlusal view.

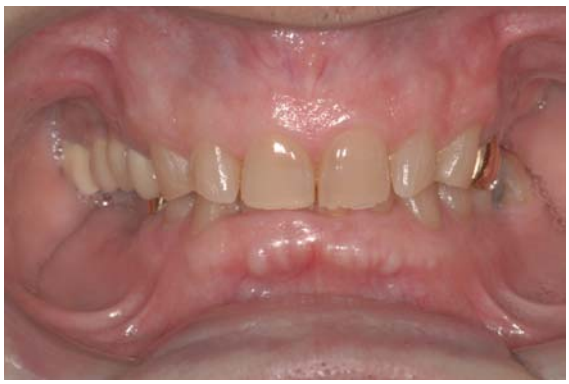

Figure 3 Preoperative frontal view.

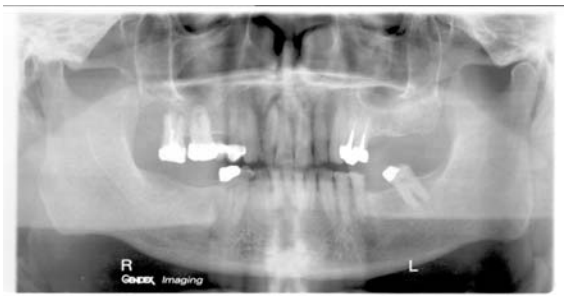

Figure 4 Preoperative panoramic x-ray.

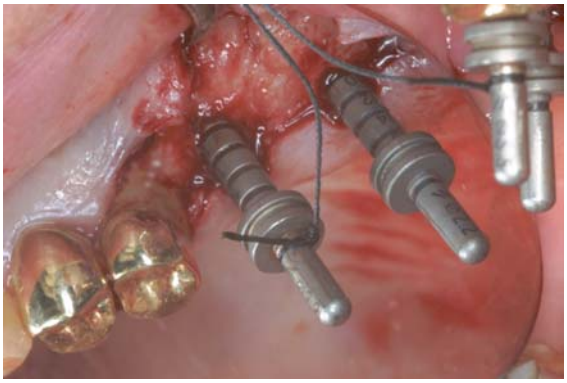

Figure 5 Drilling depth of maxillary left first and second molar implants to 10 mm.

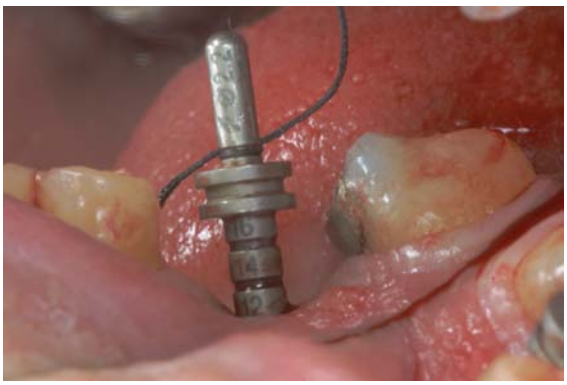

Figure 6 Drilling depth of the mandibular left first molar implant to 12 mm.

evaluated for proper healing and a panoramic radiograph was made to verify implant alignment (Fig 8).

The implants were allowed to heal for an extended period (6 months) to allow for adequate osseointegration. At this time it was determined that all implants had successfully osseointegrated. Solid WNI abutments (Straumann) were inserted and tightened to 35 Ncm using a torque controlling driver (Straumann). Abutments were adjusted clinically to ensure occlusal clearance. Abutment level impressions were made for both the maxillary and mandibular arches. Protective caps were then adjusted for proper occlusal clearance and temporarily cemented onto the abutments with eugenol-free Temp Bond (Kerr Corporation, Orange, CA).

Full veneer gold crowns were fabricated in the dental laboratory. Gold crowns were selected, because the patient described a habit of bruxism. Crowns were tried-in, fit was refined, and crowns were inserted using Ketac cement (3M ESPE, St. Paul, MN) (Figs 9–11).

The patient returned for a 1-year post-operative visit. Periapical radiographs were taken to examine for implant failure. No periapical pathosis was noted. The implants and restorations were also examined clinically for signs of failure; none were noted (Figs 12–14).

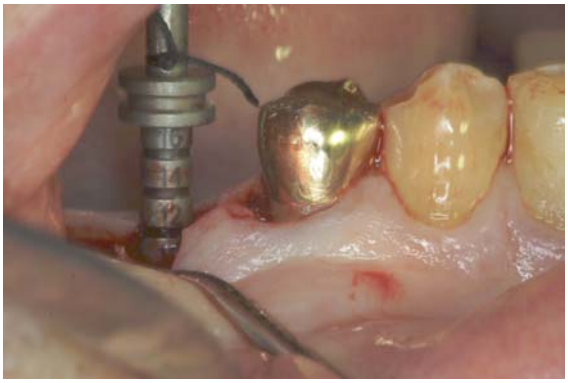

Figure 7 Drilling depth of the mandibular right first molar implant to 12 mm.

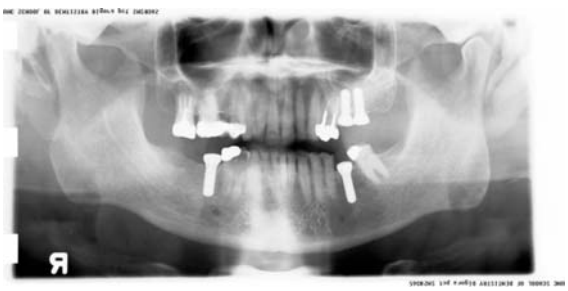

Figure 8 Implant alignment 1 week post-operative.

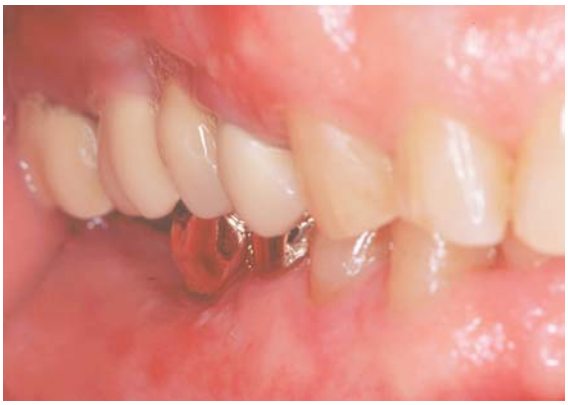

Figure 9 Final right lateral view.

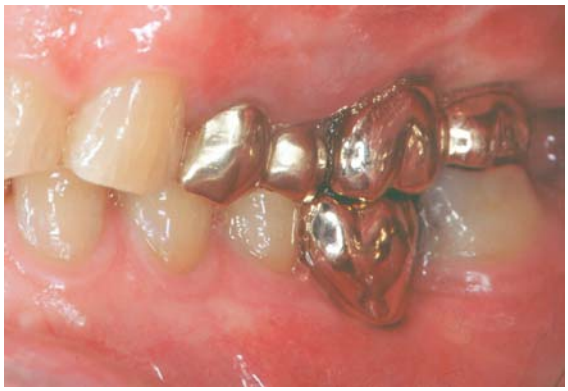

Figure 10 Final left lateral view.

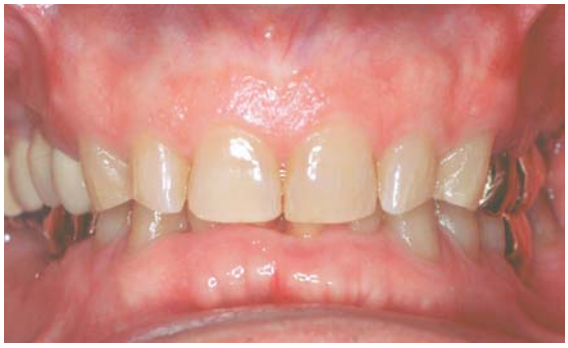

Figure 11 Postoperative frontal view.

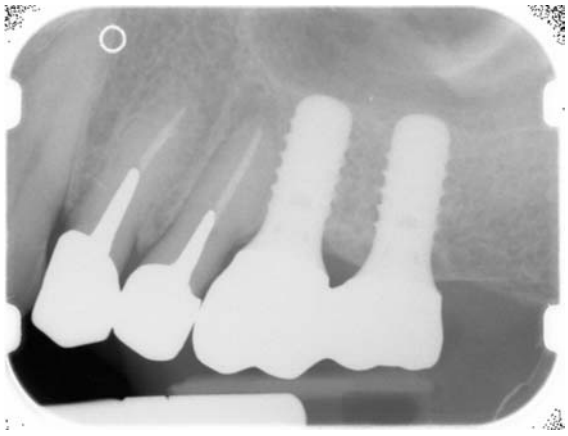

Figure 12 Maxillary left first and second molar implants 1 year post-operative.

Discussion

It is important to emphasize that the bone in the proposed implant sites was determined to be unaffected by the patient's PDB prior to the placement of the implants; for this reason it was decided to proceed with the proposed treatment plan. The decision to proceed with treatment was made by consulting with an oral radiologist and performing close examination of the preoperative radiographs, which included periapical radiographs for each site and a panoramic radiograph. Despite this preoperative assessment, clinically the bone was determined to be poor.

The patient's diminished bone quality cannot be assumed to be the result of PDB. Only a bone biopsy along with pathologic evaluation could establish the definitive diagnosis of PDB. At the very least, the finding of low bone density in multiple anatomic sites is an interesting coincidence. Although the bone did not show the traditional "cotton wool" radiographic appearance of PDB in the appositional phase, it is possible that the bone was in a different phase of the disease that could not easily be diagnosed radiographically. It is also possible that the bone was of poor quality for reasons other than the patient's PDB.

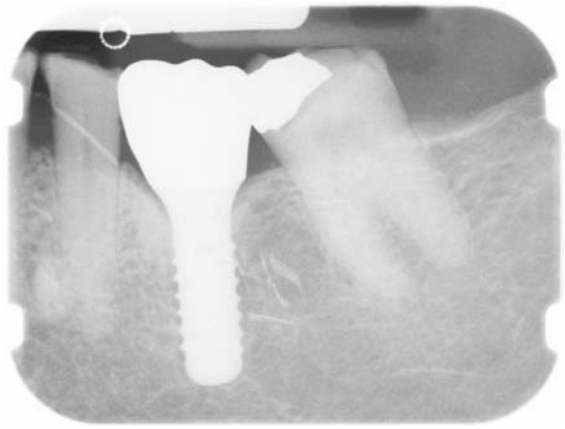

Figure 13 Mandibular left first molar implant 1 year post-operative.

If a definitive positive diagnosis of the disease had been obtained for the proposed implant sites preoperatively, and the patient's serum alkaline phosphatase concentration was at least three to four times higher than normal, limited oral bisphosphonate therapy may have been considered prior to implant placement. This drug therapy would have been considered in order to improve the bone quality in the area and to decrease the potential hypervascularity in the proposed sites. Because an oral bisphosphonate would have been considered for a relatively short period of treatment, the fear of inducing osteonecrosis of the jaw would have been minimal. Osteonecrosis of the jaw has been observed in some patients having intravenous or oral bisphosphonate therapy for cancer or osteoporosis treatment.[36–38] This treatment would have been in consultation with the patient's personal physician.

Considering the unfavorable bone density, the widest drill in the sequence was omitted in order to improve primary stability in the maxillary left first and second molars and the mandibular right first molar sites. This increase in stability was due to the implant being placed in an undersized osteotomy. The mandibular left first molar osteotomy was prepared with the complete drilling sequence. The decision to include the final drill diameter in the mandibular left first molar site was made by the surgeon in response to the increased resistance encountered during placement of the two maxillary implants with undersized osteotomies. When primary stability was not obtained with the mandibular left first molar implant, it was decided to again omit the final drill in the sequence for preparation of the mandibular right first molar site.

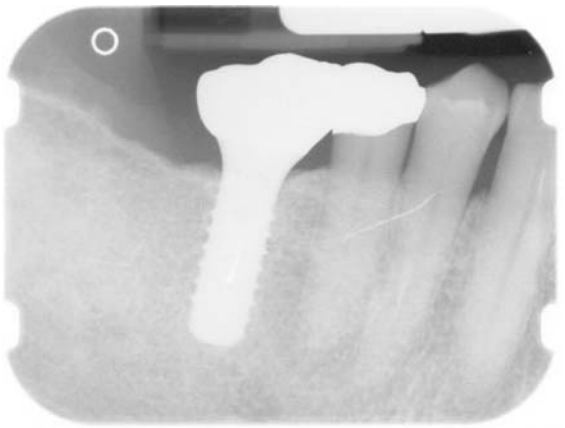

Figure 14 Mandibular right first molar implant 1 year post-operative.

Current thought is that the dental implants are contraindicated in areas affected by PDB.[39] Although PDB is commonly accepted as a contraindication to dental implants, there currently is no literature to support this rationale.[40] Even in the absence of PDB, if the quality of the bone in question is determined to be poor; implant therapy should proceed with caution or not be considered at all. On the other hand, if the bone in question is thought to be of acceptable quality, even in those patients with mild or remissive PDB, dental implants may still be a viable prosthetic consideration. If the bone is determined to be of poor quality upon clinical placement of implants, it may be possible to obtain a favorable result as seen in this particular case.

Acknowledgment

Laboratory technical work was carried out at the University of North Carolina School of Dentistry (Chapel Hill, NC) Dental Lab.

References

1. Sugarbaker ED: Osteitis deformans (Paget's disease of bones). Am J Surg 1940;48:417
2. Tiegs RD: Paget's disease of bone: indications for treatment and goals of therapy. Clin Ther 1997;19:1309-1329
3. Sharpe PT: Medical Intelligence Unit: The Molecular Biology of Paget's disease. Georgetown, TX, R.G. Landes, 1996, p 45
4. Meunier PJ, Coindre JM, Edouard CM, et al: Bone histomorphometry in Paget's disease. Arth Rheum 1980;23:1095
5. Siris ES, Canfield RE: Paget's disease of bone. in Becker KL (ed): Principles and Practice of Endocrinology and Metabolism. Philadelphia, PA, Lippincott; 1990, pp 504–512
6. Altman RD, Bloch DA, Hochberg MC, et al: Prevalence of pelvic Paget's disease of bone in the United States. J Bone Miner Res 2000;15:461–465
7. Cooper C, Schafheutle K, Dennison E, et al: The epidemiology of Paget's disease in Britain: is the prevalence decreasing? J Bon Miner Res 1999;14:192–197
8. Paget J: On a form of chronic inflammation of bone (osteitis deformans). Med Chir Trans 1877;60:37–63
9. Sofaer JA, Holloway SM, Emery AE: A family study of Paget's disease of bone. J Epidemiol Community Health 1983;37:226–231
10. Siris ES, Ottman R, Flaster E, et al: Familial aggragation of Paget's disease of bone. J Bone Miner Res 1991;6:495–500
11. Rebel A, Malkani K, Basle M, et al: Particularities ultrastructuales des osteoclstes de la maladie de Paget. Rev Rheum 1974;41:767–771
12. Mills BG, Singer FR: Nuclear inclusions in Paget's disease of Bone. Science 1976;194:201–202
13. Mii Y, Miyauchi Y, Honoki K, et al: Electron microscopic evidence of a viral nature for osteoclast inclusions in Paget's disease of bone. Virchows Arch 1994;424:99–104

14. Cartwright EJ, Gordon MT, Freemont AJ, et al: Paramyxovirus and Paget's disease. J Med Virol 1993;40:133–141
15. Barry HC: Paget's disease of Bone. London, E&S Livingstone, 1969, p 82
16. Dalinka MK, Aronchick JM, Haddad JG Jr: Paget's disease. Orthop Clin North Am 1983;14:3–19
17. Cawley MI: Complications of Paget's disease of bone. Gerontology 1983;29:276–287
18. Kaplan FS, Haddad JG, Singer FR: Paget's disease: complications and controversies. Calcif Tissue Int 1994;55: 75–78
19. Smith BJ, Eveson JW: Paget's disease of bone with particular reference to dentistry. J Oral Pathol 1981;10:233–247
20. Altman RD: Paget's disease of bone, in: Coe FL and Favus MJ (eds): Disorders of Bone and Mineral Metabolism (ed 2). Philadelphia, PA, Lippincott Williams and Wilkins, 2002, p. 1010
21. Branemark PI, Lindstrom J, Hallen O, et al: Reconstruction of the defective mandible. Scand J of Plast Reconstr Surg 1975; 9:116–128.
22. Branemark PI, Hansson BO, Adell R, et al: Osseointegrated implants in the treatment of the edentulous jaw. Experience from a 10-year period. Scand J Plastic Reconstr Surg Suppl 1977;16: 1–132
23. Adell R, Lekholm U, Rockler B, et al: A 15-year study of osseointegrated implants in the treatment of the edentulous jaw. Int J Oral Surg 1981;10:387–416
24. Albrektsson T, Branemark PI, Hansson HA et al: J. Osseointegrated titanium implants. Requirements for ensuring a long-lasting, direct bone-to-implant anchorage in man. Acta Orthop Scand 1981;52:155–170
25. Albrektsson T: Direct bone anchorage of dental implants. J Prosthet Dent 1983;50:255–261
26. Branemark PI: Osseointegration and its experimental background. J Prosthet Dent 1983;50:399–410
27. Albrektsson T: Dental implants: a review of clinical approaches. Aust Prosthodont Soc Bull 1985;15:7–25
28. Albrektsson T, Jansson T, Lekholm U: Osseointegrated dental implants. Dent Clin North Am 1986;30:151–174
29. Carlsson L, Rostlund T, Albrektsson B et al: Osseointegration of titanium implants. Acta Orthop Scand 1986;57:285–289
30. Albrektsson T, Jacobsson M: Bone-metal interface in osseointegration. J Prosthet Dent 1987;57:597–607
31. Albrektsson T, Dahl E, Enbom L, et al: Osseointegrated oral implants. A Swedish multicenter study of 8139 consecutively inserted Nobelpharma implants. J Periodon 1988;59:287–296
32. Albrektsson T, Lekholm U: Osseointegration: current state of the art. Dent Clin North Am 1989;33:537–554
33. Jaffin RA, Berman CL: The excessive loss of Branemark fixtures in type IV bone: a 5-year analysis. J Periodont 1991;62:2–4
34. Roberts WE, Simmons KE, Garetto LP, et al: Bone physiology and metabolism in dental implantology: risk factors for osteoporosis and other metabolic bone diseases. Implant Dent 1992;1:11–21
35. Uppal HS, D'Souza AR, Proops DW: Osseo-integration in Paget's disease: the bone-anchored hearing aid in the rehabilitation of Pagetic deafness. J Laryngol Otol 2001;115: 903–906
36. Munshi NC, Barlogie B, Desikan KR, et al: Novel approaches in myeloma therapy. Semin Oncol 1999;26:28–34
37. Ruggiero SL, Mehrotra B, Rosenberg TJ, et al: Osteonecrosis of the jaws associated with the use of bisphosphonates: a review of 63 cases. J Oral Maxillofac Surg 2004;62:527–534
38. Purcell PM, Boyd IW: Bisphosphonates and osteonecrosis of the jaw. Med J Aust 2005;182:417–418
39. Misch CE: Contemporary Implant Dentistry (ed 2). St. Louis, MO, Mosby, 1999, p 61
40. Medline search containing the following search topics: Paget's, Dental, Implants, Osteitis Deforms, Osseointegration