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Case Report

Complex Therapeutic Process due to Diagnostic Error in the Periapical Fibro-Osseous Lesion of Mandibular First Molar. A Case Report with Successful Implant Placement

Won-Bae Park^{1,2}, Gazelle Jean Crasto⁴, Richard Y Hong⁴, Ji-Young Han³, Philip Kang^{4*}

¹Department of Periodontology, School of Dentistry, Kyung Hee University; Seoul 02447, Korea

²Private Practice in Periodontics and Implant Dentistry, Seoul 02771, Korea

³Department of Periodontology, Division of Dentistry, Hanyang University, College of Medicine, 222-1 Wangsimni-ro, Seongdong-gu, Seoul 04763, Korea

⁴Division of Periodontics, Section of Oral, Diagnostic and Rehabilitation Sciences Columbia University College of Dental Medicine, #PH7E-110, 630 W. 168 St., New York, NY 10032, USA

*Corresponding author: Philip Kang, Division of Periodontics, Section of Oral, Diagnostic and Rehabilitation Sciences Columbia University College of Dental Medicine, #PH7E-110, 630 W. 168 St., New York, NY 10032, USA

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Abstract

Cemento-osseous dysplasia (COD) present radiographically as radiolucent lesions that are frequently misdiagnosed as endodontic lesions. The following case report involves a 42-year-old female that was treated for a molar endodontic lesion. The typically benign lesion, post endodontic therapy caused pain and chewing discomfort. The tooth was extracted a year later and biopsy samples confirmed the initial lesion was focal cemento osseous dysplasia (FCOD). The site was rehabilitated with dental implants and supplemented with bone graft material. The histological evidence of the extraction site revealed osteoporotic large bony marrow spaces with an inflammatory cell infiltrate, supplemented with cells of hematopoietic origin. Typically, sites that have abnormal bone quality, with confirmed diagnosis of cemento-osseous dysplasia are not considered ideal sites to receive implant placements. The present case report demonstrates a sequence of events for the management of FCOD in the posterior mandible with successful implant and bone graft treatment. Typically asymptomatic, FCOD benign lesions are not ideal candidates for implant placement, the following case reports depicts favorable outcomes in terms of osteointegration of the dental implant and subsequent oral rehabilitation for improved function.

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Introduction

Implant osseointegration is dependent on the dynamics of the bony tissue during both initial placement and subsequent healing. Specifically, placing implants in areas of high bone density, which is associated with increased mineral content, may result in compression necrosis [1], while areas of low bone density may compromise implant stability [2]. Similarly, dysplastic bone observed in fibro-osseous lesions presents a challenge for implant rehabilitation. The quality of available bone, in terms of structural presentations, vascular support, dense inflammatory infiltrate and the lack of cellular components directly impacts the process of osseointegration of dental implants. Cemento-osseous dysplasia (COD) is a benign fibro-osseous lesion derived from fibroblasts of periodontal ligament cells in the tooth-bearing region of the jaw [3-5]. The early osteolytic stage of COD consists of well-defined and well-vascularized fibrous tissue [6], which translates to decreased bone density often misdiagnosed in radiographs as periapical lesions of endodontic origin [5]. These lesions are observed to progress to radiopaque presentations as the bone density increases significant [7,8]. Among different classifications of COD, focal cemento-osseous dysplasia (FCOD) involves a single site, often associated with a tooth in the posterior mandible [4,9]. FCOD does not warrant any treatment [12,13]. However, early or intermediate stage FCOD are often misdiagnosed as periapical granuloma or abscess, subsequently receiving endodontic treatment. Various clinical reports document dental treatments that have led to detrimental effects due to misdiagnosis of FCOD [5,14]. The purpose of this case report is to document a sequence of events, from misdiagnosis of FCOD to successful implant rehabilitation treatment in the management of the fibro-osseous lesion in the mandibular first molar.

Case Presentation

A 42-year old female non-smoker, presents with no known systemic disease or medication regiment. Patient presented to a private dental clinic, exhibiting symptoms of pain and discomfort associated with severe tooth mobility in the mandibular right posterior region. Radiographic evaluation demonstrated alveolar bone resorption around the mandibular right first molar and periapical radiolucent lesion around distal root apex located above the mandibular canal (Figure 1a, b). The CBCT sections of the distal root demonstrate a radiopaque lesion with a radiolucent rim (Figure 1c, d). Based on the radiographic findings, the lesion

was identified and diagnosed as FCOD. The patient subsequently was referred out to an endodontist for an ailing tooth on another quadrant. The endodontist incorrectly identified, diagnosed, and treated the mandibular right first molar that was previously diagnosed as FCOD. Post-treatment, the patient reported back with persistent chewing discomfort, mobility, and pain. At this time, an apicoectomy and excisional biopsy of the periapical lesion were performed by an oral surgeon. The lesion was diagnosed as fibrous dysplasia from the histological examination of the biopsy specimen. The patient then returned to our private clinic after a one year healing period. The patient continued to report functional problems with regards to food consumption, mainly from the persistent mobility in the right mandibular molars. Patient wanted to replace the teeth with implant-supported restorations. Risks and benefits of the extraction and implant treatment were clearly outlined and discussed with the patient in detail. Another set of panoramic radiograph and CBCT were acquired for implant treatment planning purposes (Figure 2a, b). The right mandibular first molar that received the apicoectomy presented with no visible abnormalities in the area of the previous fibro-osseous lesion (Figure 2c, d).

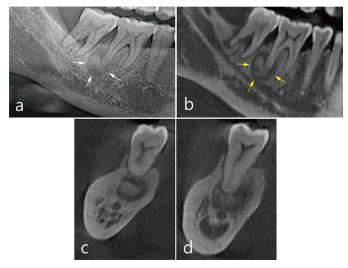


Figure 1: (a) A radiolucent periapical lesion was observed in the distal root of tooth #30 on a preoperative panoramic radiography; (b) In the sagittal image of CBCT, unilocular mixed radiolucent and radiopaque image with well-defined border was observed within the lesions. The lesion extended to the upper part of the mandibular canal; (c, d) In the coronal image of CBCT, a calcified mass was surrounded with radiolucent rim. Radiologically, the periapical lesion was suggested to be focal cemento-osseous dysplasia (FCOD).



Figure 2: Radiographic findings one year after FCOD excision with apicoectomy. (a) In the panoramic radiograph, radiolucent trabecular bone was observed around the previous DCOD site; (b) In the sagittal image of CBCT, an osteoporotic site was present at the previous FCOD site; (c) Coronal image of CBCT scanned at the distal root of #30 tooth. An osteoporotic marrow was observed at the previous FCOD site; (d) An osteoporotic area was also observed at the mesial site of #31 tooth extraction socket.

Implant Site Preparation

The right mandibular first molar had a healing period of one year after the apicoectomy and removal of the fibro-osseous lesion (Figure 3a). After local anesthesia, mucoperiosteal flap was reflected. Extraction of right mandibular first and second molars were performed and core-biopsy was acquired from the distal extraction socket of the first molar using a \emptyset 3.0 mm trephine drill (Figure 3b, c). The previous pathologic site clinically presented with bone of low density. The extraction sockets were thoroughly debrided using surgical and periodontal curettes. The osteotomy of the first molar site was prepared to ensure that the implant would be placed at the mesial root socket that was not previously

encased or associated with the fibro-osseous lesion. However, the proximity of the roots and the connected bony marrow space did not provide complete isolation of this implant from the adjacent area of previously pathology (Figure 3b). Ø4.3 x 10mm implant was placed first molar area and Ø4.8 x 8mm implant in the second molar area (Implantium, Dentium, Suwon, Korea) (Figure 3d). Particulate bone graft substitute (Osteon II, Genoss, Suwon, Korea) was placed in the peri-implant defects and covered with resorbable collagen membrane (Genoss, Suwon, Korea) (Figure 3e). Primary closure was achieved with 4-0 Nylon (Figure 3f). Antibiotics and anti-inflammatory drugs were prescribed, and patient was instructed to rinse with chlorhexidine rinse for one week.

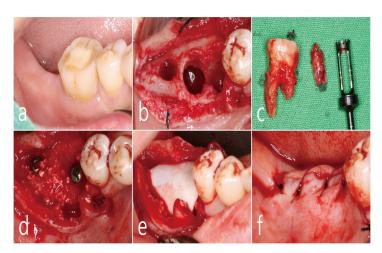


Figure 3: (a) Clinical findings 1 year after excision of FCOD; (b, c) Core-biopsy was performed on the previously excised FCOD using a Ø3mm trephine drill. Residual osteoporotic sites were thoroughly removed using periodontal and surgical curettes; (d) After the osteoporotic site around the implant was densely filled with Osteon, a Ø4.8x10mm Implant was placed; (e) The perimplant defect was covered with resorbable collagen membrane; (f) The flap was closed with 4-0 Nylon.

Micro-CT Examination

The biopsy specimen was fixed in 10% buffered formalin and micro-CT examination was performed. The scanner tube voltage was 130KV with a resolution of 14.91 μ m (intensity 60 μ A). The specimen obtained from the site of previously removed fibro-osseous lesion demonstrated large marrow spaces towards the coronal aspect with associated loose trabeculae bone surrounding the marrow space. The sample did not demonstrate any localized distinct radiopaque pattern synonymous with FCOD. No unusual abnormalities or dense bony islands were noted (Figure 4a, b).

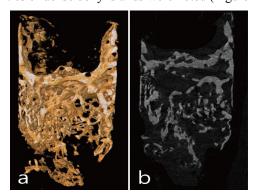


Figure 4. Micro-CT images. (a) The appearance of the specimen appears to have some bone; (b) The inside of the specimen has a very loose trabecular pattern.

Histopathological Examination

The biopsy specimen was calcified and stained with hematoxylin & eosin (H&E) (Figure 5), and the Masson's trichrome (MT) for observation of the bony trabeculae, marrow spaces and cellular infiltrate, associated collagen fibres and structural woven and remodelled bone (Figure 6). Histological analysis was performed using an optical microscope (BX-51, Olympus Optical, Tokyo, Japan). In some areas of the specimen, microscopic findings showed uniformly distributed curvilinear-shaped trabecular of woven and immature bone within proliferating fibroblastic and vascularized stroma (Figure 5a). There was no osteoblastic rimming the middle third of the specimen, a finding which would normally indicate recurrence of the originally diagnosed fibrous dysplasia (Figure 5b). The hematopoietic bony marrow space was clearly identified in the sample and encased within was dense cellular infiltrate composed of erythroid, lymphocyte, and fatty tissue (Figure 5c). An infiltrate of inflammatory cells was observed (Figure 5c), with localized clusters of multinucleated giant cells (Figure 5d). Masson's trichrome staining allowed for identification of immature bone (Figure 6a) with ginger root-shaped bony trabeculae and a fibroblastic stroma, along with curvilinear-shaped woven bone, findings that normally be seen in a COD. There was a distinct lack of osteoblastic rimming around woven bone (Figure 6b). Bone trabeculae were scarce (Figure 6c).

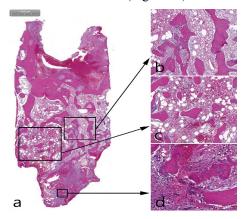


Figure 5: Core-biopsy obtained from a previous FCOD site (H&E stain). The specimen in contact with the root apex is encapsulated with soft tissue. (a) The upper part is the distal root portion resected by apicoectomy of #30 tooth, and innumerable erythrocytes and fibrous tissues were observed. Below is a previous FCOD site; (b) Microscopic finding shows uniformly distributed curvilinear-shaped trabeculae of woven/immature bone within proliferating fibroblastic and vascularized stroma; (c) A hematopoietic bone marrow containing a lot of erythrocytes, lymphocytes, and fat cells was observed. Infiltration of inflammatory cells was also observed. Woven bone exhibited lack of brush borders; (d) There was no osteoblastic rimming around woven bone in the dense fibroblastic stroma. The proliferation of fibroblasts is evident. A cluster of multinucleated giant cells was found.

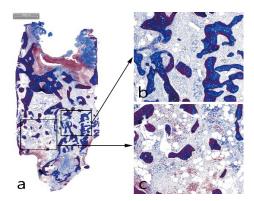


Figure 6: Core-biopsy obtained from a previous FCOD site (MT stain). (a) Only immature or woven bone exists in the specimen, and no mature bone is observed; (b) Ginger root-shaped immature bony trabeculae were observed in fibroblastic stroma. Curvilinear-shaped woven bone was found. There was lack of osteoblastic rimming around woven bone; (c) Bone trabeculae are scarce. Fat cells, erythrocytes and inflammatory cells are mainly distributed. There was no proliferation of fibroblasts in the very loose stroma.

Postoperative Management

During the follow-up visits, patient reported transient pain and swelling during the first one week post-treatment, with no other adverse events. Sutures were removed after 10 days. Implant uncovering procedure was performed after six months (Figure 7a). After reflecting the buccal flap, the cover screws were removed and healing abutments were inserted. Regenerated bone could be seen in the peri-implant defects (Figure 7b). Final prosthesis were delivery two months after placement of healing abutments (Figure 7c). Patient was recalled every 6 months. Panoramic radiograph and CBCT were taken one year after prosthesis delivery (Figure 8). The panoramic demonstrated an increase in bone density around the site of previous lesion (Figure 8a). The CBCT revealed significant increased bone density around the implant (Figure 8b). In the coronal section of CBCT, osteoporotic marrow showed increased radiopacity due to bone graft particles and bone formation (Figure 8c-d).

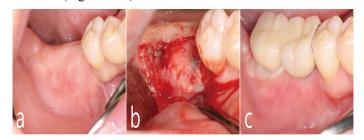


Figure 7: (a) In the clinical picture six months after implant placement, wound exposure was not observed; (b) In the uncovering procedure, peri-implant defects and lesion-related

defects were regenerated with bone tissue; (c) The prosthesis was delivered 2 months after the uncovering procedure.

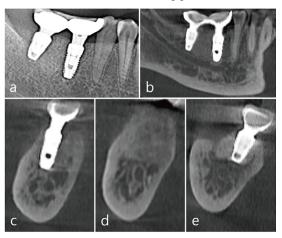


Figure 8: Radiological findings 1 year after prosthesis delivery. **(a)** On panoramic radiography, there was an increase in bone density around the previous lesion, and no recurrence was found; **(b)** In the panoramic image of CBCT, the bone density around the previous lesion and implant was significantly increased; **(c-e)** In the coronal image of CBCT, osteoporotic marrow showed increased radiopacity due to bone graft particles and bone formation.

Results

In the present case, implant rehabilitation was performed in the mandibular posterior region with a previous fibro-osseous lesion. One year after prosthesis delivery, the patient's chewing function was restored and maintained and there was no recurrence of any adverse signs and symptoms.

Discussion

This case report shows a complex oral rehabilitation process caused by a series of misdiagnosis that occurred surrounding a periapical region of mandibular first molar. The series of events lead to eventual successful therapeutic management of the site with implant placement, restoration and one year maintenance at a site of previous fibro-osseous lesion. FCOD is asymptomatic in nature and a self-limiting disease. Therefore, FCOD does not require treatment and should be monitored long-term on an annual basis [13,16,17]. However, periapical COD and FCOD are very similar in radiographic presentation to periapical infections of endodontic origin [16]. Pulp vitality tests are essential for the differential diagnosis of periapical radiolucent lesions. This is to distinguish between lesions of endodontic and non-endodontic origins [5], and to avoid unnecessary endodontic treatment [3,23,24]. The differential diagnosis of a periapical radiolucent lesion can include, but not limited to, periapical granuloma or cyst, FCOD, osteomyelitis, ossifying/cementifying fibroma, and osteoblastoma

[13,23,25]. Treatment of secondary infections or recurrence of FCOD is difficult and complex [21,22]. In present case, periapical irritation with over-instrumentation of the root canal exacerbated the situation. In the present case, the lesion was first diagnosed as fibrous dysplasia by a pathologist based on the first biopsy results. Fibrous dysplasia is a common fibro-osseous lesion that has similarities to FCOD, and in severe cases can cause cosmetic and functional disturbances [26,27]. Radiologically, fibrous dysplasia can be variably shown as sclerotic, "ground glass" appearance, and mixed radiolucent images [10,11]. However, in present case, the radiographs did not show ground-glass appearance nor expansion and perforation of buccal bone. Histologically, fibrous dysplasia shows trabeculae with prominent osteoblastic rimming while FCOD mainly shows curvilinear trabeculae ("ginger root" pattern), such as the ones seen in the histologic examination of the second biopsy specimen in the present case. Based on the clinical and radiological presentation, as well as evidence from the histologic evaluation of the second biopsy, the differential diagnosis of FCOD seems to be more accurate in the present case. compared to fibrous dysplasia or periapical lesion of endodontic origin. Implant placement in the area with FCOD is not an absolute contraindication but does come with increased risk for complications. Bone density at the site is crucial in determining implant placement, stability and eventual osseointegration [14,29]. Several reports have demonstrated that successful implant placement is possible despite the existence of COD [14,15,30,31]. However, in the early or intermediate stages of COD, the sites are typically more fibrous in nature than mineralized tissue. This puts the implant at an increased risk of failure due to the possibility of the implant being encased in soft tissue rather than bone. Clinically, early or intermediate stages of COD demonstrate multiple small fragments of fibro-osseous lesions that cannot be completely identified or removed [28]. Therefore, there is also a risk of recurrence of COD that was previously removed. Previous reports documented implant placement in the osteoporotic or hematopoietic bone marrow [15,32-25], fibrous dysplasia [36,37], and ossifying fibroma [29,38]. Thorough debridement of the sites should be performed during implant site preparation with a clear clinical assessment of bone quality and quantity. The authors recommend tailored individual care based on a complete evaluation that includes routine radiographic and clinical assessment of case history, desires and needs of the patient, the risks and benefits of implant therapy, as well as supplementing the decision tree with histological evidence of the lesion. The histological examination aids in accurate diagnosis and influences the treatment plan. With an accurate, definitive diagnosis, a long-term monitoring and care regimen can then be established.

Conclusion

Within the limitation of this case report, a definitive diagnosis

of peri-apical lesions should be clearly established prior to treatment. The patient should be made aware of the diagnosis and its implications for future complications by clear communication. For patients with fibro-osseous lesions, such as FCOD, implant rehabilitation is a possible intervention; however, each case should be individually assessed and tailored based on various phases of clinical presentation and the associated bone quality.

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