An unusual case report of peri-implant cystic lesion Loutsi Salti¹, Khaled Al-ouf²

SUMMARY

A new entity called "peri-implant cyst" has recently been described. This lesion is an inflammatory odontogenic cyst on an osseointegrated implant. It arises from inflammatory stimulation and proliferation of the rests of Malassez. Although a relatively rare condition but potentially the cause of implant failure. Peri- implant cystic lesion exhibits the same radiographic and histopathological features as the radicular cyst. The diagnosis rests on the histology, radiology, and on their correlation with clinical features, in particular the location of the lesion. Herein, we report a case of a 58-year-old female diagnosed as peri-implant cyst in the periapical area of the right maxillary first premolar implant. The microscopic examination revealed a cystic lining composed of non-keratinized stratified squamous epithelium covered with multilayered epithelium. The lesion was surgically enucleated along with preservation of the implant and wit guided bone regeneration (GBR) method, without any postoperative complications and satisfactory healing, was achieved. One year later, a newly formed bone at the apical region of the right maxillary first premolar implant was radiologically observed.

Keywords: peri-implant cyst, odontogenic cyst, peri-implant lesion, dental implant.

INTRODUCTION

Recently, a distinct pathological entity called peri-implant cyst has been introduced to describe an inflammatory odontogenic cyst on an osseointegrated implant (1). Except for its signature location at the level of the implant, the histopathological features, origin and location of the peri-implant cyst is analogous to a radicular cyst. It is a chronic inflammatory lesion that can develop several years after implant placemen due to inflammation stimulating the proliferation of epithelial rests of Malassez. (2). It is indeed often associated with inflammation and infection around the implant, potentially leading to implant failure. The pathogenesis of this cyst is not fully understood, but various contributing factors are considered, suggesting a multifactorial origin. Previous authors postulate that this cystic lesion result from trauma and infection related to dental implantation (3). This case documents a patient with a large peri-implant lesion that was found to have a central cystic cavity and an epithelial cyst lining on histopathological examination.

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CASE REPORT

In May 2019, a 58 year-old healthy, nonsmoking woman presented with mild swelling and pain in the area of the right maxillary first premolar. The patient experienced damage to their first right maxillary and second molar after endodontic treatment without crown restoration. To restore function in the right maxillary area where the two teeth were extracted, the patient opted for a four-unit porcelain bridge supported by two implants, specifically in the first premolar and second molar regions. This setup, placed in 2015, utilizes two implants, one each in the premolar and molar area, to support the bridge. She experienced the bridge repeatedly falling out. Intraoral examination revealed soft and fluctuating well-defined swelling measuring 2 cm × 2 cm in the apical region of maxillary right first premolar and extending distally to involve the right first molar region. The overlying mucosa was nonulcerated. No discharge, suppuration, and bleeding were present. The area was asymptomatic, and no sinus tract could be detected. The keratinized mucosa was inflamed (erythematous) on the facial surface of the maxillary right first premolar implant.. Oral hygiene was good. A 5-6 mm peri-implant depth was recorded circumferentially. Lymph nodes were non-palpable. Panoramic radiograph revealed welldefined periapical radiolucency in relation to right

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Fig. 1. Preoperative panoramic radiograph showing well-defined periapical radiolucency in relation to right maxillary first premolar implant (white arrow)



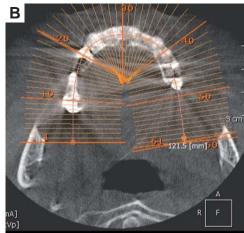




Fig. 2. CBCT scan that showed a hypodense image in the periapical region of right maxillary first premolar implant. A – sagittal, B – axial and C – coronal views.

maxillary first premolar implant (Figure 1). The patient submitted to cone beam computed tomography (CBCT) for further exploration. CBCT showed a well-defined radiolucency with well-defined sclerotic border in the periapical region of the first premolar implant, second premolar and first molar with buccal expansion, measuring 2×2.2×1.5 mm (Figure 2).

CASE MANAGEMENT

Based on the history and clinical examination as well as radiographic examination, a provisional diagnosis of peri-implant cystic lesion involving apical third of the implant was made. After removal of the bridge, stability of the implants was evaluated using the resonance frequency analysis (Osstell implant stability quotient (ISQ), Gothenburg, Sweden). Implant stability quotient of first premolar and second molar implants was recorded, respectively, 65.5 and 74.5. After administering local anesthesia, horizontal incision on the alveolar crest and two vertical releasing incisions were given on the labial aspect extending from the first maxillary premolar to second maxillary molar to reflect full thickness mucoperiosteal flap. Buccal bone expansion at the first premolar was evident. In the apical third of the implant, a 7 mm of the overlying thinned bone was removed with bur under copious irrigation to expose the cystic mass and the apical third of the first premolar implant (Figure 3). The defect did not extend to the periodontal ligament of the canine. Then, the cystic mass and all remaining tissue tags were carefully removed. Excised tissue was sent for histopatho-

logical investigation. The apical third surface of the implant surface was copiously washed with saline and chlorhexidine. The surface was then burnished with tetracycline solution (50 mg/ml) on a cotton pledged for 2 minutes. The implant remained stable. Thereafter, the defect was grafted with synthetic bone graft substitute (BONITmatrix®, 0.6×4.5 mm, Germany) mixed with autologous blood and covered with bilayer bioabsorbable barrier 20×30×0.2 mm (Hypro-Sorb® F, Czech Republic). An implant 8.5 mm in length and diameter of 3 mm was installed in the region of first molar. Borders are confronted and sutured with 4-0 silk sutures. The patient was instructed not to chew on that side or brush the surgical area for the first 4 weeks postoperatively. In the follow up period of 1 year, radiographically the bone graft became indistinguishable from the

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surrounding bone, which indicates complete bone regeneration. The missing teeth were replaced with a five-unit implant supported bridge (Figure 4). The patient was examined 12 months later, and no sign of swelling or inflammation at the side of the lesion was observed. Radiographic examination showed that the radiolucency was no longer visible, and that new bone had formed at the apical third of the implant (Figure 5).

HISTOPATHO-LOGICAL ANALYSIS

Histopathologic examination revealed a cystic structure lined by non-keratinizing squamous epithelium; this epithelium was stratified in two-three layers. The connective tissue was moderately infiltrated with lymphoid cells (Figure 6).

DISCUSSION

With peri-implant cystic lesion being relatively new pathological entity, there is little scientific evidence to suggest a cause of the cyst at

present. However, according to the recent literature such lesion is an inflammatory odontogenic cyst associated with an implant (1). Identifying the specific cause of the cysts can be challenging because they can arise from a variety of sources, including residual granulation tissue after tooth extraction and trauma (6). Under normal conditions, cell rests of Malassez remain in the form of a meshwork of cells around the root, particularly in the apical region. It has been shown that these cells persist even after tooth extraction and may be found in edentulous alveolar bone many years after extraction (7). Various inflammatory mediators present



Fig. 3. Surgical enucleation



Fig. 4. Four implant-supported fixed bridge

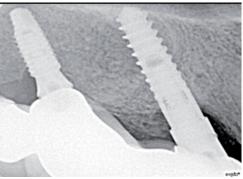
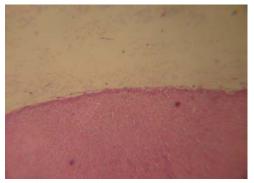




Fig. 5. Post-operative periapical radiographs, one year after the surgery, showing new formed bone at the apical region of the right maxillary first premolar implant



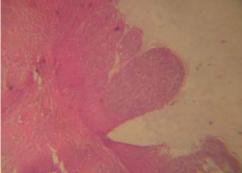


Fig. 6. Histopathology picture. The epithelial lining of the cyst lumen is visible in H&E stain (original magnification $\times 10$)

in the area around the tooth apex can stimulate the epithelial cell rests of Malassez to proliferate and form the cyst's epithelial lining (8). Surgical implantation in the osteotomy prior to or with insertion of the implant has been described as an alternative source of epithelium that might proliferate into a cystic lesion around a titanium implant. Frisch and Screaton suggested that displaced mucosal cells lose their proliferation capacity and undergo apoptosis when separated from connective tissue (9). Another possible source of epithelium for the development of peri implant cyst may be from a residual sinus lining around the pre-existing tooth (10, 11). In the

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case described and three reviewed reports, the peri implant cystic lesion was frequently found around the long implants in the apical region (1, 6, 12). Rests of Malassez, as noted earlier, persist more frequent around the apical third of the root. As a consequence, any persisting inflammatory lesion is likely to be in the apical region. Because of this, peri - implant lesions tend to affect long implants placed in dense bone (13). In the present case, the peri-implant cyst was large. It has been reported that the growth rate of a radicular cyst in the primary dentition is approximately 4 mm each year (14). Accordingly, initial formation of the cyst was more likely 4 years after implantation. In order to lower the risk of peri-implant biological complications, a minimum recall interval of 5-6 months has been recommended (15). In our patient, the implants were not followed. The patient could not attend follow up and control visits. In the present case, the right maxillary first premolar and the right maxillary first molar regions had D4 bone. Hence, we kept the first premolar implant in its place to maintain the bone. Based on patient's financial status and implant stability value of the first premolar, we have decided to keep the right maxillary first premolar implant and placed one additional implant. After informing the patient about potential implant failure risk factors, an additional implant has been placed in the first molar region to increase the implant surface area. Increasing implant surface area enhances restoration retention and reduce the risk of overload to the first premolar implant (16, 17). Considering bone density

in posterior maxilla, insertion of additional implant will compensate for less dense bone and can help distribute occlusal forces more evenly (17). Been studied clinically and radiographically, guided bone regeneration (GBR) technique is a successful procedure employed to aid reparation process of destroyed periapical tissues after surgical elimination of the periapical pathology (18). Furthermore, GBR is a generally predictable method technique for regenerating bone around exposed implant (19). The clinical and histopathological features of our case could suggest that the potential etiopathogenesis was either presence of periapical bone pathology before implant placement or contamination of the apical part of the implant. To the best of our knowledge, this case is very unique and rare, since the periimplant cystic lesion was large.

CONCLUSION

Peri-implant cyst is a rare pathology that affects the apical third region of an implant. It has a negative effect on the long-term outcome of implant therapy. This case report emphasizes the importance of follow-up in implant therapy. Furthermore, it highlights the successful healing of a large peri-implant cystic lesion through enucleation of the cyst, filling the resulting cavity with synthetic bone graft substitute mixed with autologous blood, and covering the area with bilayer bioabsorbable barrier. Healing was uneventful for the involved implant, and no mobility and displacement was present at 1-year follow-up.

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