Treatment of massive infected residual cyst with marsupialization, decompression, surgical endodontic therapy, and enucleation: Case report

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ABSTRACT

Management of residual cyst by marsupialization, decompression, surgical endodontic therapy, is reported here. Though typically surgical enucleation is the recommended treatment for small cystic lesions, Treatment of large lesions may require a treatment plan that is delivered through two stages: first stage is marsupialization and decompression. Second stage is surgical endodontic treatment accompanied by curettage and surgical enucleation. A case report is presented of a massive residual dental cyst. Presentation, diagnosis and management of the massive cyst are discussed.

Keywords: Residual cyst, marsupialization, Cyst decompression, surgical endodontic therapy.

1. INTRODUCTION

Odontogenic cyst of the jaws rarely grows into extensively large lesions, when they do develop; they are commonly arising from odontogenic Keratocyst or dentigerous cyst. Residual cysts are often discovered during routine dental radiographs. They are typically asymptomatic unless infected (Dimitroulis and Curtin, 1998). Residual dental cysts are an apically located inflammatory periodontal cyst that persists after extraction of the associated tooth (Main, 1970; Killey et al., 1977). These cysts arise from the proliferation of epithelial cells rests of Malassez in the periodontal ligament that are stimulated by ingress of inflammatory mediators originating from necrotic pulp of the associated tooth. The development of radicular cysts has been attributed to the continuation of this inflammatory process. Upon extraction of the associated tooth the majority of radicular cysts regress, however, a small percentage of these lesions remains static or become infected and grow (Dimitroulis and Curtin, 1998; Main, 1970; Oehlers, 1970). Most of odontogenic jaw cysts are found in the anterior maxillary area followed by lower premolar area (Nair,
Prevalence of residual cysts reported in literature varies according to sampling type and methods of investigation. In a radiographic study reviewing the panoramic radiographs of asymptomatic patients presented for routine dental visit it was reported at 2% (Perrelet et al., 19776). In another study where histological examination was carried out on lesions enucleated from the jaws, residual cysts were reported at 7.6% (Main, 1970). Of all odontogenic dental cysts, it is believed that residual dental cysts compromise 10% of those odontogenic dental cysts (Main, 1970; Killey et al., 1977). A study by Oehlers et al., (1970) followed the progression of residual cysts. In a radiographic examination of 168 cases that were presumed to be residual cysts, 92.3% of those lesions resolved within 9 months of extraction while 7.7% remainstatic. No cases showed increase in size of lesion. Although this study attempts to report the percentage of radicular cysts transforming into residual cysts, the pre-operative diagnosis of those cases as cystic lesions were based on radiographic interpretation rather than histological findings. If a residual cyst becomes infected, this will lead to the development of signs and symptoms which are typically treated by enucleation of the lesion. Sukegawa et al., (2015) published a case report of a primary intraosseous squamous cell carcinoma possibly arising from infected residual cyst; however, other than this case no other reported cases of residual cyst malignant transformation have been seen. Yet it is crucial to identify cases of infected residual bone cyst and provides necessary treatment as the continued growth of these lesions may lead to destruction of neighboring structures and affects patients’ overall health.

2. THE CASE

A 40-year-old male patient presented to the endodontic department with a chief complains of a swelling inside my mouth that has been there for over a year from 5 August 2021 to 31 January 2022. His dentist referred him to our clinic to have a root canal treatment on left maxillary canine. The patient presented to his dentist with a palatal swelling over a year ago. Root canal treatment was done on maxillary left lateral incisor. Treatment was followed by periapical surgery few months after due to persistent swelling and finally extraction of the tooth was done 2 months ago. Patient was referred to our office for root canal treatment on left maxillary canine when the palatal swelling did not subside after extraction. Detailed medical history was taken. Patient is healthy with no history of any systemic diseases or abnormalities.

Patient states that he is a social smoker and denies any allergies to any medications. Vital signs were recorded BP 125/82 mmHg, Pulse rate 74 BPM, Respiratory rate 12 breaths/minute, Oral temperature 96.8°F. Upon examination Patient is alert, oriented with no distress. Extra oral exam reveals no noticeable swelling or facial asymmetry. No lymphadenopathy or extra oral sinus tract. Intra oral exam shows a large fluctuant swelling in hard palate sized 3.5 cm by 2.5 cm that extends from mesial of left maxillary central incisor to mesial of left maxillary second premolar (figure 1). The swelling did not cross the midline. No sinus tract is detected. Oral cancer screening is negative. No shift of uvula from midline. Patient has poor oral hygiene with multiple missing teeth, heavily restored teeth, and generalized mild marginal gingivitis. Maxillary left central incisor has mesial and distal composite fillings with discolored margins and a palatal amalgam filling. Maxillary left lateral incisor and first premolar are missing. Maxillary left canin has a small palatal composite filling with intact margins. Maxillary left second premolar has a composite build up with an open distal margin. On palpation a depression on buccal attached gingiva in the area between maxillary left central incisor and maxillary left canin was noticed suggesting bone fenestration. Clinical testing was done, and results presented in Table 1.

Figure 1 (A) and (B) Pre-operative intraoral views of the patient demonstrating a palatal swelling of 3.5 x 2.5 cm at the left maxilla.
Aspirational biopsy was performed. A suppurative exudate was aspirated mixed with blood and tissue fluid. A total of 6 PA radiographs were taken for maxillary first incisor, canin, and second premolar of total 2 PA radiograph per tooth (Figure 2 A-F). All three teeth have defective fillings. There is a localized horizontal bone loss in the area of maxillary lateral incisor. A large well defined radiolucent lesion is seen in the anterior maxilla that measures approx. 2 cm by 3.5 cm. The lesion is centered around maxillary canin, but it extends from root of maxillary central incisor to distal of maxillary canin. There is periapical radiolucency around root second premolar that is 4 mm by 2 mm, separate from the radiolucent lesion in the anterior maxilla. Root of second premolar has an apical curvature towards the mesial. Maxillary first and second molars are heavily restored with normal crestal bone, intact lamina dura and bone trabeculation around their roots.

Table 1 Clinical testing

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Figure 2 Pre-operative periapical radiographs (A-D) showing a large well-defined radiolucent lesion in the apical region of maxillary left central incisor and centered around maxillary canin. E and F showing pre-operative periapical radiograph of maxillary posterior teeth demonstrating a periapical radiolucency of approximately 2 X 4 mm around roots of second premolar.

CBCT scan was taken for the maxilla. It shows a well-defined radiolucency of approximately 2cm by 3 cm in the anterior left maxilla. The lesion extends from maxillary central incisor to maxillary second premolar. It appears rounded with thinning/erosion/destruction of both buccal and palatal cortical plates. Expansion of cortical bone with associated intraoral swelling was indicators that the lesion was benign. The nasal floor has been displaced superiorly. Small periapical pathology is also noted at the
apices of second premolar, separate from the radiolucency in the anterior maxilla. Sinus mucositis is observed. Selected axial, coronal and sagittal views (figure 3). Maxillary left central incisor was diagnosed as having a necrotic pulp with acute apical abscess while maxillary second premolar was diagnosed with pulpal necrosis and acute apical periodontitis. First and second molars are diagnosed as having normal pulp with normal apical tissues. Due to the extensive size of the periradicular lesion involving the entire left premaxilla with loss of both buccal and palatal bone performing a root canal treatment followed by surgical enucleation would involve and disrupt the vascular supply, neural structures on neighboring vital structures. It might lead to devitalization of neighboring teeth, development of soft tissue defects and damage to adjacent anatomical structures. It was decided that the best course of treatment would be performing a non surgical root canal treatment for maxillary left central incisor and maxillary second premolar followed by decompression and irrigation to allow progressive reduction in lesion size and minimize the complication of surgical enucleation which would be performed later after the lesion has decreased in size.

After reviewing patient medical history, discussing pros and cons of treatment, treatment options and obtaining a consent non surgical root canal treatment of maxillary left central incisor was initiated. Buccal and palatal infiltrations were given using 2 cartridges of 1.7 ml of 2% lidocaine with 1:100,000 epinephrine followed by rubber dam isolation of tooth of interest. No fractures were detected on inspection. A purulent exudate was visualized in the canal and was allowed to drain. Working length established with apex locator. Cleaning and shaping were done. 20 ml irrigation with 6% NaOCl throughout canal instrumentation was used once working length was confirmed. Irrigation with 3 ml of 17% EDTA was done followed by final irrigation with 6% NaOCl. Canal was dried then obturation was done following vertical compaction technique. Access restored with Glass Ionomer placed. PA radiograph was taken (Figure 4) and checked the occlusion. Post operative instruction given. Patient was referred back to the general dentist for restoration to stabilize the tooth. My discussion with the general dentist included arriving at a definitive restorative treatment plan after we were sure the endodontic care was complete.

**Figure 3** A) Axial, B) Curved sagittal, C) Coronal sections of Computerized Tomography (CT scan) of Maxillary teeth showing well-defined radiolucent lesion of 4X3 cm in diameter with thinning / destruction of buccal and palatal cortical plate around left central incisor.
On the following visit non-surgical root canal treatment was performed for maxillary second premolar following same protocol described earlier. Post-operative radiograph were shown in figure (5). Patient came for follow-up approximately 3 months following the root canal treatment of maxillary central incisor. Palatal swelling still presents with little reduction in size (figure 6). Patient reports no pain or discomfort. Pulp testing was repeated on maxillary left canin with no change in findings. Marsupialization along with irrigation was suggested to the patient to allow for drainage, reduce the pressure, and minimize complications following surgical enucleation of the lesion. Informed consent was obtained. Buccal infiltration was given using one cartridge of 1.7 mL of lidocaine with 1:100,000 of epinephrine at the area of missing maxillary left lateral incisor. A small incision was made through an area in the gingiva overlying a bone fenestration, creating a round window in attached gingiva, underlying periosteum and bone of approx. 1 cm in diameter. This was followed by dissection, removal of granulation tissue and inspection of the lesion. The lesion was found to involve palatal cortical bone. Initial impression of the lesion was that it is a cystic lesion. 20 ml of sterile saline irrigation was used through the created window. Margins of the cystic tissue lining were sutured to the attached gingiva in the margins of the created window. Penrose drain placed and sutured in place using 6-0 black silk sutures.
Figure 6 Intraoral views of the patient taken 3 months after Root Canal Treatment demonstrating persistence of palatal swelling.

Patient was given irrigation needles and instructed to irrigate through the drain with salt water 2-3 times a day. Patient was prescribed Ibuprofen 600 mg to be used every 4-6 hours for 3 days. Post operative instructions were given. The patient was contacted by phone on the night of surgery. He reported minimum discomfort, controlled by medication and denies any excessive bleeding or adverse reaction to the procedure. Patient came for follow up ten days later, reporting a decrease in size of the intraoral swelling and denies any pain or discomfort. He states that he has been irrigating through the penrose drain 3-4 times a day. Upon examination, penrose drain still in place with minimal inflammation around sutures. Palatal swelling was decreased significantly. Patient was advised to continue irrigating the lesion through the drain at least twice a day. Though there was a reduction in swelling, and signs of healing, several factors remained: the lesion is believed to be cystic, its critical size, and the loss of both buccal and cortical bone. Enucleation of the lesion with placement of resorbable membrane and bone graft along with periapical surgery of maxillary left central incisor was determined to be the optimal treatment. Patient was scheduled for 2-month appointment for evaluation, CBCT and possible periapical surgery of maxillary left central incisor and enucleation. Patient came 10 weeks later, for follow up and surgical root canal treatment of maxillary central incisor and enucleation of the palatal lesion.

A PA radiograph and CBCT were taken and showed significant bone healing and reduction in size of periapical radiolucency (Figure 7). Discussed pros and cons of treatment and treatment options; informed consent was obtained. Buccal and palatal infiltration was given using 2 cartridges of 1.7 ml of 2% lidocaine with 1:100,000 epinephrine and 1 cartridge of 2% lidocaine with 1:50,000 epinephrine in order to achieve proper anesthesia and hemostasis. Drain was removed. A triangular full thickness mucoperiosteal flap design was chosen. Reflection of flap revealed perforation of the cortical bone overlying the periapical lesion. Osteotomy window was enlarged to allow complete enucleation of the lesion. A clinical picture was taken before and after the enlargement. The bulk of the lesion was curetted, stored in formalin and sent for histological examination. The lesion seemed to extend from mesial of maxillary left central incisor to distal surface of maxillary left canin.
The clinical impression of the lesion was that it is cystic. The apical 3 mm of the root of maxillary left central incisor was resected. After resection the root surface was stained with methylene blue and inspected for fracture, cracks or missed canals. Retro-prep of main canal was done for a depth of 3 mm. The area was then washed with saline. Retro-filling was done using Bioceram root end filling material. A resorbable collagen membrane was placed in the crypt and packed at the area of the palatal cortical plate perforation, followed by 2 CCs of allograft bone then another membenewas placed overlying the bone graft at the area of buccal bone perforation and sutured to periosteum (Figure 8). The flap was then placed back and sutured with 6-0 black silk suture. 3 interrupted sutures placed for interproximal areas and horizontal locked mattress suture for the vertical incision. Additionally, interrupted sutures were placed in the area where penrose drain was placed. A PA radiograph was taken (Figure 9). Patient received ice packs and verbal and written postoperative instructions. Patient was prescribed Vicodine 5/500 to be used every 4-6 hours for 3 days, Ibuprofen 600 mg to be used every 4-6 hours for 3 days, and 1 bottle of Peridex mouth wash to be used twice a day for 1-2 weeks. Patient was contacted by phone on the night of surgery. He reported moderatediscomfort controlled by medication and denies any excessive bleeding or adverseraction to theprocedure.

Patient presented 5 days post-op for suture removal. Patient was asymptomatic. Gingival healing was uneventful. No signs of infection or gingival necrosis or inflammation. Sutures were removed. Patient was seen for follow up 6 weeks after surgery. There were no recurrences of signs or symptoms. Gingival tissues retained positive architecture, healthy gingival color and gingival stippling. No signs of inflammation, infection or pus formation. Teeth involved in the surgical site remained vital and normally responding to pulp vitality testing. No recession around teeth involved in the surgical site. Clinical pictures were taken showing resolution of palatal swelling (Figure 10). Long term follow up was not possible as the patient moved out of the country permanently. The patient was treated in King Abdulaziz University. Consent was signed by the patient to undergo dental treatment and use images and radiographs in any future scientific publication in accordance with HIPPA and King Abdulaziz University regulations. Treatment was completed in 5.5 months.

**Figure 7** Follow up (A) Periapical radiograph and (B) CT scan of maxillary teeth showing signs of bone healing and reduction in the size of periapical radiolucency.
Figure 8 Clinical and radiographic pictures of apical surgery of maxillary left central incisor during apical surgery. (A) Osteotomy window after enlargement following flab reflection. (B) Periapical radiograph was taken following resection, retro-prep and retro-fill to verify the angle of resection, adequate depth of retro-prep and adequate condensation of retro-fill. (C) Resorbable collagen membrane was placed at the palatal depth of the defect. (D) 2 CC of allograft bone was packed against the membrane. (E) Resorbable collagen membrane was placed at the buccal aspect of the defect and sutured to periosteum.
Figure 9 Immediate Post-operative Periapical radiograph of maxillary left central incisor following surgery.

Figure 10 (A) and (B) Post-operative intraoral views of the patient taken 6 weeks after surgery showing resolution of palatal swelling up and healing of gingival tissues.

3. DISCUSSION

Maxillofacial region is one on the most common sites for the development of odontogenic cysts as well as neoplastic lesion. Differentiating them based solely on clinical or radiographic examination can be challenging (Nair et al., 2008). A cyst is defined as a pathological cavity with a defined wall of connective tissue and an epithelial lining, filled with liquid, semiliquid or gaseous content. Growth of a cyst is typically slow, centrifugal and infiltrative (Asián-González et al., 2007). The proliferations of epithelial cells of Malassez in the periodontal ligament are attributed to the rise of Residual cysts. These proliferative activities of epithelial cells are stimulated by ingress of inflammatory mediators originating from necrotic pulp of the associated tooth. This inflammatory process leads to the development of radicular cyst. Upon extraction of the associated tooth the majorities of radicular cysts regress, however, a small percentage of these lesions remain static or become infected and grow (Dimitroulis and Curtin, 1998; Main, 1970; Oehler, 1970).

The treatment of large cystic lesions is an area of prolonged debate (Gallego-Romero et al., 2002). Reports on management procedures for large periapical lesions range from conventional nonsurgical root canal treatment with long-term calcium hydroxide therapy to various surgical interventions. The recommended treatment for small radicular cyst is total enucleation. For larger cysts
marsupialization followed by surgical enucleation is the best approach (Freedl and 1997; Neaverth and Burg 1982). Performing marsupialization prior to surgical enucleation will lead to reduction of the size of the lesion which will minimize damage to vital structures, avoid devitalization of neighboring teeth, simplify surgical procedure, lower operative complication and improve postoperative healing (Martin, 2007). This is in agreement of de Moraes et al., (2020) study where they demonstrated marsupialization prior to total enucleation of a large Calcifying odontogenic cyst proved to be a favorable and effective treatment choice. Following this surgical technique resulted in a significant reduction of the lesion, minimal damage to anatomical structures was and promoted bone repair. This approach minimized the overall morbidity and costs; it removed the need for a surgical reconstruction following enucleation and resulted in an excellent outcome.

In a case report by Torres-Lagares et al., (2011) a large maxillary cyst was treated for 3 months with marsupialization and decompression, followed by surgical endodontic therapy of the affected teeth and cystectomy. Nonsurgical endodontic therapy alone will typically lead to the healing of small cystic lesion. With larger cystic lesions additional treatment procedures are usually necessary to ensure complete healing. If surgical enucleation of a large cystic lesion was performed as a solo procedure without a prior marsupialization and decompression for prolonged period, a damage of neighboring teeth or anatomic structures will be encountered. To avoid this sequel, treatment should begin with the more conservative approach of decompression, to reduce the size of the lesion, followed by a surgical procedure of apicoectomy and cystectomy. In this case they reported complete resolution of the lesion after 8 months of surgical enucleation.

AL-Omar and Elmorsy (2017) reported that, a gradual reduction in the radiolucent area was apparent after 6 months of marsupialization and decompression, and cyst enucleation was performed easily and good stability in central incisor. Resolution of the lesion was complete after 8 months. In a retrospective study conducted by Demir and Günhan (2021), they compared the treatment outcome and effectiveness of only enucleation, enucleation with platelet rich plasma (PRP) application and marsupialization in management of dentigerous cyst. They reported best treatment outcome in terms of bone regeneration was observed with PRP application with no difference in bone healing when enucleation and marsupialization were performed. Similar findings were reported by Riachi and Tabarani (2010) in which they compared healing in 2 cases of radicular cysts treated by either by surgical enucleation or by marsupialization. They demonstrated complete healing and conservation of vital structures using either aggressive or minimal invasive approach.

Yun et al., (2014) advocated the use of marsupialization or decompression technique treating cystic lesions among young patients. This technique has been successfully performed in growing children and adolescents, demonstrated a successful reduction in size, with minimal intraoperative complications with minimal and did not interfere with eruption of permanent teeth. Successful treatment of large cysts using initial marsupialization and a second phase surgery with enucleation and curettage has been previously reported.

4. CONCLUSION

Cystic lesions that are presented with large dimensions, as seen in the present case, a conservative approach of delivering a two-stage treatment is recommended. Following a 2-stage approach with minimize the damage to important structures eliminates the need for aggressive and expensive surgical reconstruction. This clinical case is in agreement with the findings of previous authors who believed in the viability of the procedure. In the case reported here, a substantial reduction in the radiolucent area was apparent after 10 weeks of marsupialization and decompression, and cystectomy was performed easily and good stability in central incisor.

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Author Contributions
The author designed the treatment plan for the patient, delivered treatment, and conducted the literature research, manuscript preparation, editing and review.

Ethical approval
Ethical approved cleared by ethic committee of King Abdulaziz University, Faculty of Dentistry, Department of Endodontics,
Informed consent
Written & Oral informed consent was obtained from the participant included in the study.

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Conflicts of interest
The authors declare that there are no conflicts of interests.

Data and materials availability
All data associated with this study are present in the paper.

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