UNUSUALLY LARGE RADICULAR CYSTS OF MAXILLA: STEPS IN DIAGNOSIS & REVIEW OF MANAGEMENT

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ABSTRACT

Radicular cysts are common odontogenic cyst of jaw bone which invariably presents as well-defined radiolucency around the apex of the involved tooth. It is diagnosed on routine radiography or causes a slow growing swelling, limited to apical region of a tooth. Radicular cyst presenting as unusually large maxillary lesions is uncommon. This paper reports two cases of large radicular cyst involving maxilla. The first case presented as a single lesion occupying almost the entire anterior maxilla, while the second case presented a large cyst occupying the entire left half of the maxilla and the maxillary sinus. Unusually large radicular cyst of maxilla require stepwise clinical, radiological and laboratory evaluation to reach a definitive diagnosis. Although no consensus exist regarding the precise treatment modality for large radicular cyst of maxilla, both cases were successfully treated by surgical removal of the cyst following endodontic therapy.

Keywords: Large, Radicular cyst, Maxilla, Enucleation

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INTRODUCTION
Radicular Cysts are believed to be formed from epithelial cell rests of Malassez (ERM), which are remnants of Hertwig’s epithelial root sheath, present within the periodontal ligament. Proliferation of these epithelial cell rests is frequently associated with stimuli from periapical periodontal inflammation secondary to pulpitis (1). During periapical inflammation, host cells in the periapical tissues release many inflammatory mediators, proinflammatory cytokines, and growth factors which induce proliferation of the ERM in all directions to form a three-dimensional ball mass(2,3). As the epithelial mass grows, the central cells move further away from their source of nutrition and undergo necrosis and liquefaction degeneration, forming central cystic cavity lined by epithelial wall. Following its formation, radicular cysts grows by periapical bone resorption mediated by prostaglandins and cytokines. It is interesting to note that most inflammatory mediators which induce proliferation of epithelial cell rests also mediate bone resorption in inflammatory periapical lesion (4-9). However there is no time-lapse study showing that the size of apical cysts gradually increases as periapical bone destruction increases. Clinically radicular cyst presents as periapical lesion, ranging from 0.5 to 1.5 cm in size. Smaller periapical cysts can often be treated conservatively by non-surgical endodontic therapy. Proper endodontic therapy of the involved teeth removes irritants in the canals by chemomechanical instrumentation. As the root canal is completely sealed, all cell components participating in inflammatory reaction gradually resolve (10). Subsequently majority of inflammatory cells, endothelial cells and fibroblasts in the paeriapical lesion are deleted by apoptosis or programmed cell death (11). However surgical removal following endodontic treatment is required for cases not amenable to conservative therapy (12). Presence of unusually large periapical lesion may often lead to diagnostic dilemma. Such cases need to be evaluated in a step wise manner to reach a definite diagnosis. This article reports two cases of large radicular cyst of maxilla thepresentation, steps in diagnosis and management of these lesions are also discussed.

Case Report:
Case 1:A 34 year old female presented with complaint of painless swelling over roof of the mouth. The patient first noticed the swelling about 6 months back, which had grown gradually to its present size. No history of trauma or tooth pain was reported. Extraoral examination revealed no facial swelling or obvious facial asymmetry. Intraoral examination showed a palatal swelling starting distal to right 1st premolar and extending anteriorly, across the midline to the contra lateral incisor, measuring about 4 x 3.5 cm in size (Figure 1a). Swelling was non-tender and firm on palpation. The overlying palatal mucosa appeared normal. Maxillary anterior teeth were grade one mobile, with normal depth of gingival sulcus on diagnostic probing. All the maxillary
antlers were non-vital on electric pulp testing. The clinical presentation was suggestive of a benign intraosseous tumor or an odontogenic cyst. Clinical differential diagnosis included central giant cell lesion, radicular cyst, odontogenic Keratocyst, globulomaxillary cyst and ameloblastoma. Aspiration of the lesion yielded yellow color fluid which was subjected to cytochemical evaluation (Figure 1b). The fluid consisted of cholesterol crystal and numerous inflammatory cells, suggestive of an inflammatory cystic lesion.

A cone beam computed tomography (sironaorthophos X3GD, Germany) was advised, to study the size and extent of the lesion. The 3D reconstruction showed a large osteolytic lesion extending form maxillary right 1st premolar to maxillary left canine and superiorly till the floor of the nasal fossa. The roots of the associated teeth were displaced and showed no resorption (Figure 2a). Axial section showed 5 x 3.5 cm intraosseous radiolucent lesion with defined sclerotic borders involving almost entire anterior maxilla (Figure 2b). Diagnosis of inflammatory odontogenic cyst was established based on the clinical, radiological and aspirate evaluation. Surgical enucleation of the cyst was planned under general anesthesia after root canal therapy of the involved maxillary teeth. Biomechanical perpetration and irrigation (with saline & 1% sodium hypochlorite alternatively) of root canals of all the maxillary anterior was done under antibiotic cover. Calcium hydroxide intracanal medication was placed for period of 1 week. Subsequently the root canals were obturated with gutta-percha. The cyst was approached through labial gingiva mucoperiosteal flap and removed by enucleation. Apicoectomy and retrograde root end filling for maxillary anterior teeth was done with MTA to obtain apical seal. The labial and interdental bone support around the maxillary anterior teeth appeared to be compromised due to bone resorption caused by the periapical lesion. After through irrigation of the bone cavity & hemostasis, the bone defect was grafted with platelet rich concentrate to facilitate periapical wound healing. Autologous platelet rich concentrate was prepared from 10 ml of patient’s own blood centrifuged at 2888 rpm for 12 minutes (Remi Lab centrifuge) (Figure 3). The defect was closed primarily and cyst lining was sent for histopathology examination, which confirmed the diagnosis of radicular cyst [Figure 4]. Splinting of maxillary anterior teeth was done for 3 weeks, using 22-gauge stainless steel wire stabilized with light cured composite in immediate postoperative period. At 1 year follow up the patient was asymptomatic and showed no sign of recurrence. There was no mobility of the anterior teeth and imaging of the surgical site showed normal healing of the bone defect (Figure 5).

Case 2:A thirty six year old female, presented with complain of painless swelling of eight months duration over left side of midface. The patient gave history of fall from bicycle during her childhood. On examination a large extraoral swelling was evident, extending from left infraorbital region to level of corner of mouth, measuring roughly 4 x 5 cm in size. The nasolabial fold was obliterated along
with elevation of ala of left nose (Figure 6). The swelling was non-tender and firm on palpation. Intraoral examination showed soft fluctuant swelling localized over left maxillary vestibule in premolar-molar region. Maxillary left central and later incisors were non-responsive to electric pulp vitality test. On radiological evaluation, a large radiolucent lesion was evident involving the entire left maxilla. Orthopantamogram showed a well-defined lesion with sclerotic border, extending from maxillary midline till the maxillary tuberosity. The entire left maxillary sinus appeared hazy on paranasal sinus view (Figure 7). The presenting features were suggestive of cyst of odontogenic or maxillary sinus origin. Content of the lesion was aspirated intra-orally under local anaesthesia using 18 gauge needle. Cytological examination of the straw color aspirate showed numerous inflammatory cells and cholesterol crystals. Based on the history, clinical presentation, vitality test, radiographic features and evaluation of aspirate, diagnosis of inflammatory odontogenic cyst following pulp necrosis of left maxillary incisors was made. However, looking into the unusually large size of the lesion, incision biopsy was planned to conform the diagnosis. The lesion was accessed intraorally through Caldwell-Luc approach (by creating bony window in region of canine fossa). A portion of the cyst lining was removed and sent for sent for histopathological examination. Diagnosis of radicular cyst was confirmed based on incision biopsy report. Following chemo-mechanical preparation of pulp chambers of the involved left maxillary incisors and placement of calcium hydroxide intracanal dressing for one week, the patient was prepared for surgical removal of the cyst under general anesthesia. After the cyst was enucleated in-toto, apicoectomy of the left maxillary incisors and retrograde root end filling with MTA was done. The cyst lining was submitted for microscopic evaluation which confirmed diagnosis of radicular cyst (Figure 8). The postoperative healing was uneventful. The patient was symptom free and showed healing intrasosseous surgical defect, at 6 months follow-up. Radicular cysts are the most common odontogenic cyst affecting the human jaw (13). It is believed to be formed from proliferation of epithelial residues in the periodontal ligament as a result of periapical inflammation secondary to pulpitis. Although they are direct sequel to apical periodontitis, not every case of apical periodontitis causes formation of radicular cyst (14). Since radicular cyst form at the apex of tooth root(s) it is also called periapical cyst. They may be associated with any tooth. However it commonly involves maxillary anterior teeth and seldom involves primary dentition. Their prevalence is highest in the third decade of life and are more commonly seen in males than females (15). Position of the maxillary anterior teeth makes them more prone to injury and subsequent pulp necrosis. This may be the possible explanation for their prevalence in males and in maxillary anterior region. One of our reported cases (case 2) gave history of trauma to maxillary anterior teeth, which could have been the possible cause of

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initiation of cyst formation. Periapical inflammation and pulpitis secondary to dental caries is another cause of formation of radicular cyst. Most of the radicular cysts are symptomless and discovered when periapical radiographs are taken for non-vital teeth. They present as round to oval radiolucency associated with root apices, varying in size from 0.5 to 1.5 cm. As the cyst grows in size by resorption of the surrounding periapical bone, it causes localized jaw swelling. Pain and pus discharge are presenting feature of an infected radicular cyst. Only few cases of radicular cysts involving large part of maxilla have been previously reported in literature (16-19). Interestingly the reported cases of large radicular cyst involve maxilla and very often extended to occupy large portions of the maxillary sinus. A logical reasoning to explain this association can be the relatively thin and cancellous nature of the maxillary bone as compared to dense mandible, which would be more resistant to expansion and resorption. Also extension of these periapical lesions into the maxillary sinus would allow its unrestricted growth, leading to formation of giant radicular cyst.

Although intra-oral radiographs are sufficient for studying a periapical cysts, larger lesions often require more extensive imaging to evaluate the location, size, proximity to vital structures and extent of the lesion (20). Both extra-oral radiographs (including orthopantamogram & paranasal sinus view) and three dimensional imaging using cone beam computed tomography were used in our cases for radiological assessment. Aspiration of the cyst fluid is another important diagnostic test for cystic lesion. It helps to rule out any vascular lesion and provides fluid specimen for cytochemical evaluation. In our reported cases the cyst content was aspirated under local anaesthesia, using wide gauge needle. The fluid was yellowish-white in color and contained cholesterol crystal and numerous inflammatory cells. The treatment of radicular cyst, as a disease of root canal infection or inflammation, consists of eradicating or substantially reducing the microbial load and inflammatory mediators from the canals and periapical tissue. Numerous measures have been described to reduce the number of microorganisms in the root canal system, including the use of various irrigation regimens and intracanal medicaments. Various root canal irrigants used include Sodium hypochlorite, ethylenediaminetetraacetic acid (EDTA), citric acid and chlorhexidine. Sodium hypochloride is the most commonly used endodontic irrigant (21). In our cases 1% Sodium hypochloride and saline was used alternatively as root canal irrigant. Intracanal medicaments eliminate or reduce number of microorganisms, rendering canal contents inert. It helps in prevention of post-treatment pain, and enhances anesthesia (22). Calcium hydroxide is most widely used intracanal medicament. It also plays a major role as an inter-visit dressing in the disinfection of the root canal system. Reduction of inflammatory cells and mediators from roots of the involve tooth may often lead to resolution of smaller periapical lesion. However cases which do not respond to conservative management
require surgical intervention. Surgical treatment is preferred approach to treat large radicular cyst. For apical radiolucency larger than 20 mm in diameter or having cross-sectional area greater than 200 mm², surgical removal may be the best option (23). Surgical treatment is also recommended when the canal appears calcified or obstructed and cannot be negotiated with endodontic instruments. The surgical treatments for radicular cyst include total enucleation, marsupialization or a combination of these techniques (24, 25). Removal of entire cyst allows histopathological evaluation of cyst lining, thereby helping in reaching a definitive diagnosis and ruling out any malignant or dysplastic changes in large cyst. After the enucleation of the cyst, apicoectomy of the involve teeth is done. Apicoectomy involves sectioning of the apex of the root and retrograde filling to attain a sound periapical seal. Historically amalgam was the most frequently used material in apicoectomy procedure. IRM, super EBA and MTA are newer and more suitable materials, and give better results in apicoectomy procedures than Amalgam (26). Advantages of MTA include high biocompatibility and excellent sealing ability with low marginal leakage. However high cost of MTA is a major disadvantage to its use in every clinical situation. All our cases were treated with surgical enucleation of the periapical cyst along with apicoectomy and retrograde fillings of involved roots with MTA.

The bony defect left after the removal of the cyst lining heal uneventfully after a proper apical seal is attained and the intrasosseous bony cavity is irrigated generously to remove all periapical irritants. In our first case report we had grafted the bony cavity with autologous platelet rich concentrate to facilitate healing of the large defect of anterior maxilla, which had caused significant bone loss around the maxillary anterior teeth. The teeth were subsequently splinted in immediate postoperative period. Platelet rich concentrate is known to initiate osteoinduction process which is mediated by growth factors present in platelets. High number of platelets in the concentrate produces large amount of growth factors, initiating bone formation (27). Beside bone regeneration, it also aid in healing of surrounding soft tissues (28). Both the reported cases were followed up for 6-12 months and showed uneventful healing of the lesion with no reoccurrence or complication. Periapical pathology secondary to pulp inflammation is a common finding. However usually large periapical lesion require stepwise clinical, radiological and laboratory evaluation to reach a definitive diagnosis. Such large lesion often warrants detailed radiological assessment to study the location, size and extent of the lesion. Cytochemical evaluation of the aspirate provides valuable input regarding nature of the cyst. Although no consensus exist regarding the precise treatment modality for large radicular cyst of maxilla, both cases were successfully treated by surgical removal of the cyst following endodontic therapy of the involved teeth. Enucleation of the cyst provided the lining for complete histological evaluation of the periapical
pathology and to rule out any malignant or dysplastic changes in these unusually large cysts.

**Figure 1:** (Case 1) Swelling over anterior palate (block arrows)

![Image of swelling over anterior palate]

**Figure 2:** (Case 1) (a) Cone beam 3D reconstruction showing large radiolucent lesion involving almost entire anterior maxilla (b) Axial section, at the level of Apical third of maxillary anterior teeth showing the dimensions of the cyst.

![Image of cone beam 3D reconstruction and axial section]

**Figure 3:** (Case 1) Platelet rich concentrate prepared from patients' blood (centrifuged 10 ml blood at 2888 rpm for 12 minutes; Remi Lab centrifuge)

![Image of platelet rich concentrate preparation]
**Figure 4:** (Case 1) H & E stained Photomicrograph showing (a) Stratified squamous lining epithelium (4X magnification) (b) Cholestrol clefts (black arrow) and giant cells (red arrow) in the wall of the cyst lining (40X magnification).

![Figure 4](image)

**Figure 5:** (Case 1) One year post-operative radiological showing healing at the surgical site.

![Figure 5](image)

**Figure 6:** (Case 2) (a) Frontal (b) Lateral (c) Bird view of the extraoral swelling evident over left side of mid-face; (d) Intraoral swelling in the left maxillary vestibule

![Figure 6](image)
Figure 7: (Case 2) (a) Orthopantamogram showing large radiolucent lesion with sclerotic borders (red arrows) occupying the entire left maxilla (b) Paranasal sinus view showing haziness of the left maxillary sinus (blue arrow), as compared to the normal radiolucency of right maxillary sinus (red arrow).

Figure 8: (Case 2) (a) Cyst removed in toto (b) Photomicrograph showing (H & E stained; 10X magnification) showing cyst lining made up of stratified squamous epithelium (black arrow).
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