Management of true lateral infected radicular cyst as well as associated mucosal fenestration that occurred postoperatively through an interdisciplinary approach: A rare case report

Sanjeev Kumar Salaria, Shashank Arora, Shefali Goyal,1 Amit Khunger

Abstract:
A radicular cyst (RC) is one of the types of inflammatory cyst of odontogenic origin. It is usually associated with nonvital teeth and is mostly observed in males. It is routinely treated by nonsurgical endodontic therapy; however, if the cystic lesion does not respond to endodontic therapy, then surgical intervention with or without regeneration is the treatment of choice, depending upon the size, location of lesion, the bone integrity of the cystic lesion wall, proximity to vital structures and residual bone defect elicit after enucleation, etc., We hereby present successful interdisciplinary management of rare true lateral RC in the interproximal site with respect to teeth number 21 and 22 as well as associated mucosal fenestration that occurred postoperatively other than its apparent etiology and diagnosis.

Key words:
Enucleation, fenestration, lateral radicular cyst, regeneration, root canal treatment

INTRODUCTION
Different odontogenic cysts (OCs) are encountered in dental practice. Radicular cyst (RC) is the one of the most prevalent lesions affecting the jaws ranging from 52.2%1 to 68%,2 usually located in the anterior maxilla at the apical end of nonvital teeth; however, sometimes, it is observed on the lateral aspect of root in relation to the lateral accessory canals.3 Multiple treatments ranging from nonsurgical endodontic therapy (NSET), surgical enucleation, marsupialization, regeneration of residual hard tissue defect management following enucleation of RC, etc., have been recommended in the literature. Here, we present a successful endodontic periodontics interventional management of rare true RC lesion in the interproximal site and enucleation-associated postoperative soft tissue complication, in addition to its conceivable etiology and diagnosis.

CASE REPORT
A 20-year-old otherwise healthy female patient was reported to the institutional department of endodontics with the chief complaint of pain and swelling in the left upper front tooth for the past 2 weeks. Her history was suggestive of trauma to the face about 7 years back. On intraoral clinical examination, soft tissue swelling between tooth number (#) 21 and 22, mesial tilt of crown of #22 [Figure 1a], and tenderness on percussion of #22 were observed. #22 was reported to be nonvital whereas #21 was vital, on vitality examination. Intraoral periapical and occlusal X-ray showed large pear-shaped unilocular radiolucency on the lateral root aspect of #21, 22 with sclerotic borders [Figure 1b and c, respectively]. The patient was referred to the department of periodontology for consultation; on clinical evaluation, periodontal probing depth...
was 2-3 mm with respect to (w.r.t.) #21, 22 as well as in rest of the dentition. Fine needle aspiration performed for swelling w.r.t. #21,22 showed blood mixed yellow color fluid which, on Papanicolaou, smear revealed intermixed inflammatory cells predominately neutrophil and lymphocytes and with numerous red blood cells (RBCs), suggestive of chronic inflammatory lesion. On the basis of above, clinical findings including fluid aspirate analysis provisional diagnosis of RC was made. The patient was advised to proceed for root canal treatment (RCT); if the lesion does not respond to endodontic therapy, then surgical intervention may be planned. Root canal preparation was completed as per the standard endodontic protocol w.r.t. #22; however, clinically, the swelling remained static, and even on radiographical evaluation, the lesion did not show any changes till preobturation w.r.t #22 [Figure 2a and b, respectively] suggested that cyst is of self-sustaining type (true RC). Therefore, the patient was informed in detail regarding single stage management of lesion by root canal obturation w.r.t. #22 followed by surgical enucleation of lesion’s associated anticipated residual bone defect (RBD) management by amalgamation of platelet-rich fibrin (PRF), demineralized freeze-dried bone allograft (DFDBA) and OstIN™ bone graft with its pros and cons in comparison to the non-graft technique. The patient submitted signed consent in support of periodontist advice.

After Phase I therapy, all routine investigations advised were reported to be within normal perimeter. Root canal obturation was done in #22 utilizing lateral condensation technique. Immediately, under local anesthesia, crevicular [Figure 3a] and vertical releasing incision were given w.r.t #21–23 and mesial line angle of #21, respectively, followed by full-thickness flap reflection [Figure 3b]. Following blunt dissection till the apical extent of the lesion in the vestibule, complete enucleation of the cyst (≥15 mm × 18 mm) was carried out with the help of surgical curettes [Figure 3c]. Enucleated cystic growth was preserved in 10% formalin solution and sent for histological evaluation. RBD was thoroughly debrided [Figure 3d]; adjacent root surfaces were planned followed by the placement of amalgamation of PRF, DFDBA, and OstIN™ bone graft [Figure 3e]. During the surgical separation of cystic lining from the labial flap, mucosal perforation occurred near mucogingival junction that was approximated, and flap was sutured with 4-0 silk and radiovisual graph taken w.r.t #21 and 22 [Figure 3f] showed RBD duly filled with bone graft material. Postoperative instructions were given to the patient, and the patient reported on the 3rd week for the postoperative checkup.

Hematoxylin and eosin-stained microphotograph section under × 10 showed that cystic cavity lined by the nonkeratinized epithelium of variable thickness with arcadian pattern. In some area pseudostratified hyperplastic epithelium with underlining neutrophilic infiltration was observed. Underlying connective tissue stroma was moderately collagenous with numerous small endothelial cells line blood vessels with engorged RBCs, suggestive of RC [Figure 3g].

Surgical site healed uneventfully, sutures were removed, but mucosal fenestration was observed w.r.t #22 [Figure 3h] on 3 weeks postsurgical follow-up. The patient was informed regarding the same and signed consent was taken to repair the same with PRF in conjunction with coronally advanced mucosal flap (CAF). Under local anesthesia,fenestration site was thoroughly refreshed followed by irrigation with normal saline. Autologous Choukroun’s PRF was prepared. Partial thickness incision was given with releasing incision w.r.t #22 for tension-free displacement of trapezoidal CAF. PRF was secured at defect site and CAF was secured utilizing 4-0 vicryl suture [Figures 3i and 4a-c, respectively]. Postsurgical instructions were given to the patient. Eight days postoperatively, mucosal fenestration site healed uneventfully; at 3-month postoperatively, complete repair of mucosal fenestration was observed which remain static till 9-month postoperative [Figure 4d-f, respectively]. RBD regeneration reevaluated on radiovisual graphs at 3-month postsurgically showed radiopacity in the treated RBD site due to the stability of grafted materials and was distinguished from adjacent bone site [Figure 4g]. Complete elimination of radiopacity of the grafted materials was observed because of uniform homogenization (uniform radiopacity) of healed treated RBD, which was not distinguishable from the adjacent bone at 9-month postoperatively [Figure 4h]. No complication was observed till the last follow-up.
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DISCUSSION

RC is routinely observed in dentulous site. It usually arises from epithelial cell rests of Malassez existing within the periodontal ligament in response to necrosed pulp/truma-induced inflammation.[1,3] The incidence of RC’s peak frequency was observed in the third decade of life,[1,4] and a large number of cases were reported in the fourth and fifth decade of life,[1] with male predominance.[1,4] It is usually asymptomatic, but pain and infections are the other clinical features. The true RC did not respond to the conventional endodontic therapy.[4] Similar clinical findings were observed in the present case of RC, which was confirmed histologically too; however, it was rare because (i) only a small fraction (<10%) of

Figure 3: (a-i) Crevicular incision with respect to number 21–23; releasing incision with flap reflection; cyst enucleation; residual bone defect; graft placement; flap sutured; histology showed cyst lumen and epithelium having arcading pattern; fenestration and releasing incisions in mucosal fenestration for coronally advanced mucosal flap, respectively

Figure 4: (a-h) Flap advancement; platelet-rich fibrin placement; closure of flap; 8 days, 3- and 9-month postoperative clinical evaluation showed closure of fenestration; 3-month postoperative intraoral periapical X-ray showed graft stability and defect resolution at 9 months
the periapical lesions advanced into true radicular cysts,\textsuperscript{[5]} (ii) reported in a 20-year-old female, and (iii) to the best of our information received from web of literature, no true RC was reported in the interproximal bone site of the upper anterior teeth till date.

Conceivable etiology of RC in the present case was a history of trauma that might lead to pulpal necrosis, which may serve as stimulus for epithelial residues in the periodontal ligament, resulting in cyst formation.

It was differentially diagnosed from odontogenic keratocyst because of its aggressive nature which was discarded on the behalf of chronicity, occlusal radiographic findings, as well as its proximity to the defect.\textsuperscript{[6]} Traumatic bone cyst (TBC) commonly found in the metaphysis of long bones, while relatively rare in the jaws, representing approximately 1\% of all jaw cysts, lesions are mainly situated in the body and ramus of the mandible, and rarely in the maxilla, teeth exhibit no mobility or displacement and remain vital.\textsuperscript{[7]} Dentigerous cysts are always associated with an impacted tooth.\textsuperscript{[8]} Lateral periodontal cyst is an unusual cyst of odontogenic origin, most frequently encountered in the mandible between the roots of canines and premolars; histology demonstrates a cystic cavity supported by fibrous connective tissue and is lined by a thin nonproliferative layer of epithelium showing areas of focal thickening which may be interpersed with glycogen-containing clear cells.\textsuperscript{[9]} Nasopalatine duct cyst is the most common non-OC of the maxilla, arises from embryogenic remnants of nasopalatine duct, and usually develops in the midline of anterior maxilla near the incisive foramen, whereas histological presentation shows cystic lining composed of squamous, columnar, cuboidal, or some combination of these epithelial types with stratification.\textsuperscript{[10]} As none of the above finding exists in the present case, RC appears in the nonvital tooth.\textsuperscript{[11]} Therefore, provisional diagnosis of RC was reached on the basis of clinical, radiographical, and fluid aspirate analysis, which was later on confirmed histologically too.

A cystic cavity with complete epithelialization but no opening into the apical foramen and root canal is reported as true RC; due to independent nature, it did not heal by NSET.\textsuperscript{[12]} Such lesions may be successfully managed by extraction of the associated nonvital teeth and curettage, surgical decompression, marsupialization or enucleation, etc.\textsuperscript{[8]} Usually tissues have the proficiency to reinstate their unique structural assembly after trauma or insult through repair process which is closely synchronized through cell-cell and cell-matrix signaling via a wide variety of cytokines and growth factors, as well as by cell mitosis and apoptosis, so that the wounded tissue can be reinstate systematically.\textsuperscript{[11]} However, the damaged periapical tissues can be repaired but they will never completely return to their original structure after nonsurgical endodontic therapy (NSET) because of uncoupling reaction.\textsuperscript{[12]} Therefore, keeping in mind the age of patient, healing of periradicular tissue after NSET and self-sustaining nature of the true RC surgical enucleation followed by amalgamation of PRF, DFDBA, and ostIN™-assisted regeneration of RBD was planned and executed immediately after root canal obturation to accomplish the best outcome.

PRF, DFDBA, and ostIN™ due to their distinguishing properties and harmonized effect, such as PRF, are immaculate platelet fibrin network without any chemical adulteration, which effectively promote neovascularization, boost wound closing, fast cicatrization tissue restoration, and play substantial role in soft and hard tissue regeneration.\textsuperscript{[13,14]} DFDBA is osteoconductive in nature and acts as a source of osteoinductive factors; as it elicits mesenchymal cell migration, attachment, and osteogenesis,\textsuperscript{[15]} whereas ostIN™ (60\% synthetic hydroxyapatite and 40\% tricalcium phosphate) due to their osteoconductivity and high bioactivity reported that it improves both quality and quantity of new bone formation,\textsuperscript{[16]} it might be responsible for the excellent clinical and radiographical outcome in the present report.

The outcome achieved in the present case was in accordance with the report of Camargo \textit{et al.} and Salaria \textit{et al.}, but former had utilized the combination of autologous bone graft, Bio-Oss with PRF in the management of large periapical RC lesion following endodontic therapy,\textsuperscript{[17]} whereas the later one utilized coronally advanced flap in conjunction with PRF-assisted immediate management of residual soft tissue defect following surgical excision of pyogenic granuloma,\textsuperscript{[18]} amalgamation of PRF and hydroxyapatite bioactive glass composite granules for the management of localized advance loss of periodontal support-associated grade II fucation, and intrabony defect in chronic periodontitis patient,\textsuperscript{[19]} respectively.

**CONCLUSION**

True RC is best managed by RCT with immediate enucleation of lesion followed by amalgamation of autograft, allograft, and alloplast graft-assisted regeneration of RBD to improve the clinical and radiographical outcome as well as to reduce the patient visits and financial burden too.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Acknowledgement**

We are thankful to the management of Surendera Dental College and Research Institute, Sri Ganga Nagar, for providing all kind of instruments, equipment and technical expertise facilities needed for the case management.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**REFERENCES**

1. Shear M, Speight P. Radicular cyst and residual cyst. In: Shear M, Speight P, editors. Cysts of the Oral and Maxillofacial Regions. 4th ed. Oxford: Blackwell Munksgaard; 2007. p. 123-42.

2. Killey HC, Kay LW, Seward GR. Benign Cystic Lesions of the Jaws, Their Diagnosis and Treatment. 3rd ed. Edinburgh and London: Churchill Livingston; 1997.
3. Narula H, Ahuja B, Yeluri R, Baliga S, Munshi AK. Conservative non-surgical management of an infected radicular cyst. Contemp Clin Dent 2011;2:368-71.

4. Bhaskar SN. Oral surgery-oral pathology conference No. 17, Walter Reed Army Medical Center. Periapical lesions-types, incidence, and clinical features. Oral Surg Oral Med Oral Pathol 1966;21:657-71.

5. Nair PN. Non microbial etiology: Periapical cysts sustain post-treatment apical periodontitis. Endodontic Topics 2003;6:96-113.

6. Salaria SK, Gulati M, Ahuja S, Goyal S. Periodontal regenerative management of residual tunnel osseous defect results from the enucleation of lateral periodontal cyst in anterior maxilla: A rare case report. J Indian Soc Periodontol 2016;20:638-42.

7. Titsinides S, Kalyvas D. Traumatic bone cyst of the jaw: a case report and review of previous studies. J Dent Health Oral Disord Ther 2016;5:318-25.

8. Deshmukh J, Shrivastava R, Bharath KP, Mallikarjuna R. Giant radicular cyst of the maxilla. BMJ Case Rep 2014;2014. DOI: 10.1136/bcr-2014-203678.

9. Nart J, Gagari E, Kahn MA, Griffin TJ. Use of guided tissue regeneration in the treatment of a lateral periodontal cyst with a 7-month reentry. J Periodontol 2007;78:1560-4.

10. Dedhia P, Dedhia S, Dhokar A, Desai A. Nasopalatine duct cyst. Case Rep Dent 2013;2013:869516. doi:10.1155/2013/869516.

11. Majno G, Jorits I. editors. Cell, Tissues, and Disease. 2nd edn. Oxford: Oxford University Press; 2004. p.210 –9.

12. Seltzer S, Krasner P. editors. Endodontology: Biologic considerations in endodontic procedures. 2nd edn. Philadelphia, PA: Lea & Febiger; 1988.

13. Salaria SK, Kaur S, Sharma I, Ramalingam K. Coronally advanced flap in conjunction with platelet-rich fibrin-assisted immediate management of residual gingival defect following surgical excision of recurrent pyogenic granuloma in the maxillary esthetic segment. J Indian Soc Periodontol 2018;22:273-6.

14. Salaria SK, Ghuman SK, Kumar S, Sharma G. Management of localized advance loss of periodontal support associated Grade II furcation and intrabony defect in chronic periodontitis patient through amalgamation of platelet-rich fibrin and hydroxyapatite bioactive glass composite granules. Contemp Clin Dent 2016;7:405-8.

15. Grover V, Kapoor A, Malhotra R, Sachdeva S. Bone allografts: A review of safety and efficacy. Indian J Dent Res 2011;22:496.

16. Lee JH, Ryu MY, Baek HR, Lee KM, Seo JH, Lee HK. Fabrication and evaluation of porous beta-tricalcium phosphate/hydroxyapatite (60/40) composite as a bone graft extender using rat calvarial bone defect model. ScientificWorldJournal 2013;2013:481789.

17. Camargo JM, Braga T, Camargo R, Love RM, Reher P. Surgical management of a large radicular cyst using modified guided tissue regeneration techniques: A case report with 4 years follow-up. Oral Surgery 2019;12:323-31.