Bilaterally symmetrical infected radicular cysts: Case report and review of literature

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\section*{A B S T R A C T}
Radicular Cysts (RCs) are the most common inflammatory cysts arising from the odontogenic remnants. It is usually associated with carious, non-vital, or fractured tooth. Although the occurrence of RCs is not uncommon, but bilaterally symmetrical presentation of these cysts in young patient is quite rare. Present case was treated successfully with endodontic treatment of the adjoining involved teeth, followed by extraction of the offending teeth and enucleation of the cysts. Hereby we report case of bilaterally infected RCs in a 14 years old female patient. Also an attempt has been made to discuss its pathogenesis and clinical features in light of current information from the literature.

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\section*{1. Introduction}
The Radicular Cysts (RCs) are the most common inflammatory cysts of the oral and maxillofacial region. It arises from the epithelial remnants in the periodontal ligament following necrosis of the pulp [1]. In adulthood, RCs are the most common cystic lesions found at the apices of the teeth [2,3] and represent 52–68% of all cysts found in jaws [4]. RCs can occur at any age, however, they appear to be rare in the primary dentition, in children, representing only 0.5–3.3% [5]. Killey et al. [6] in their series reported that the incidence of RC was 68% and in the series of Jones et al. [7] it represented 63.3% of all odontogenic cysts. The age distribution showed very few cases in first decade and peak frequency in the third decade. There are a large number of cases in the fourth and fifth decades, after which there is a gradual decline [1]. The lower incidence of RCs associated with deciduous teeth has been shown in a number of studies despite high incidence of dental caries. This feature is attributable to the few epithelial cell rests resulting from the development of primary teeth [5]. The purpose of this paper is to present a case of bilateral symmetrical RCs in a young female patient and to discuss its pathogenesis and clinical features.

\section*{2. Case report}
A 14 years old female presented with a chief complaint of mild pain and swelling on the right side of the lower jaw of two months duration. There was no difficulty in chewing and swallowing. Mild halitosis and pus discharge was noticed in that area. The medical history was non-contributory. The extra-oral examination was remarkable for a 3 × 2 cm firm, localized, and tender swelling over the right body of mandible. Mouth opening was adequate. The intra-oral examination was remarkable for a localized swelling on the right side, due to the expansion of buccal cortical plate extending from canine to second molar. There was no expansion of lingual cortical plate in this region. Mandibular right first molar was broken down and only the roots were present. Oral hygiene was poor. Mandibular right first and second premolars and second molar were non-vital, and exhibited tenderness to percussion. Mandibular right second molar exhibited grade I mobility. Further examination revealed expansion of buccal cortical plate in the region between mandibular canine and second molar on the left side. Mandibular left first molar was also badly broken down with remaining roots but first and second premolars and second molar were non-vital and did not exhibit tenderness to percussion. Rest of the dentition was normal. Panoramic radiograph (OPG) showed well-defined, corticated, bilateral oval radiolucent areas in the body of the mandible related to both mandibular first molars. The radiolucent area extended anteroposteriorly to involve mandibular first and second premolars and second molars, bilaterally. The rad-

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ograph also showed resorption and loss of lamina dura of the apices of roots of mandibular second premolars and second molars bilaterally (Fig. 1).

Provisional diagnosis of infected bilateral RCs was made. Differential diagnosis of Odontogenic keratocystic tumour, Unicystic ameloblastoma, Central giant cell granuloma and Cherubism were considered.

The treatment plan included endodontic treatment of #37, #35, #34, #44, #45 and #47 under local anaesthesia, followed by extraction of #36 and #46, apicectomies of the endodontically treated teeth, and enucleation of both the cysts with primary closure, under general anaesthesia. Endodontic treatment of mandibular first and second premolars and second molars bilaterally was carried out prior to the surgery. The extraction of #36 and #46, apicectomies of the endodontically treated teeth, and enucleation of both the cysts with primary closure, was carried out under general anaesthesia.

The pathological tissues from both the lesions were sent for histological examination. The recovery was uneventful. The histological report was consistent with that of a RCs (Figs. 2 and 3). The patient was given a temporary partial denture. The patient was followed up for two years post-operatively without any complications (Fig. 4).

Fig. 1. Pre-operative OPG showing well-defined bilateral radiolucency involving remaining roots of #46 & #36. The teeth #47, #45 and #35 & #37 are showing resorption of the roots.

Fig. 2. H & E, 10X, photomicrograph showing a non-keratinized stratified squamous lining.

Fig. 3. H & E, 40X, photomicrograph showing cholesterol clefts in the connective tissue wall, characteristic of inflammatory cysts.

Fig. 4. Postoperative OPG three years after the surgery showing good healing and no recurrence.

3. Discussion

RCs are the most common inflammatory cysts arising from the epithelial residues as a result of periapical periodontitis following necrosis of the pulp. It occurs in all tooth-bearing areas of the jaws and 60% are found in the maxilla and 40% in the mandible [1]. Its size rarely exceeds 1 cm with higher incidence in the maxillary anterior region [1]. Hill reported that cysts in the primary dentition develop at a rate of 4 mm per year [8,9]. Sprawson proposed that the short physiological life of the deciduous tooth might account for the lower prevalence in the deciduous dentition [10].

The pathogenesis of RCs may be considered in three phases: phase of initiation, phase of cyst formation and phase of enlargement.

3.1. Phase of initiation

The origin of the cystic epithelium lies with cell rests of Malassez, which are the epithelial remnants of Epithelial Root Sheath of Her- twig. The epithelial cell rests are initiated to proliferate by inflammation as a result of necrotic debris and bacterial antigens/endotoxins derived from the necrotic pulp. These endotoxins have a direct effect on the epithelial proliferation, and also initiate an inflammatory response resulting in production of cytokines with pro-inflammatory and bone-resorbing activities. The major cytokines identified are IL-1...
and IL-6, and also have a direct effect on epithelial proliferation [11]. Both humoral and cell mediated reactions have been implicated in the pathogenesis. Infiltrates of T lymphocytes, indicating that cellular immune reactions are involved in their pathogenesis, were demonstrated in human periapical granulomas, by Skau et al. [12].

3.2. Phase of cyst formation

The next phase in the pathogenesis of a RCs is the process by which a cavity comes to be lined by the proliferating odontogenic epithelium. One concept proposes that the epithelium proliferates and covers the bare connective tissue surface of an abscess cavity or a cavity which may occur as a result of connective tissue breakdown by proteolytic enzyme activity [13]. The other theory, and perhaps more widely supported theory, postulates that a cyst cavity forms within a proliferating epithelial mass in an apical granuloma by degeneration and death of cells in the centre [1].

3.3. Enlargement

Toller's studies provided evidence for the hypothesis that osmosis makes a contribution to the increase in the size of cysts. The hydrostatic pressure is considered to be of primary importance in the growth of all cyst types. Kubota et al. [14] confirmed the early results of Harris and Toller [15] and Skaug, [16] that the pressure was greater than the local blood pressure. They concluded that increased pressure played a pivotal part in the early cyst growth. Glycosaminoglycans present in the walls and fluids of odontogenic cysts [17] probably contributes to the expansile growth of the cyst.

Harris et al. [18] confirmed the role of prostaglandins, their bone resorbing capacity in the enlargement of jaw cysts. Eventually, as the epithelial cell mass enlarges, the central cells become distant from their blood supply, undergoes necrosis, and breaks down, thereby forming a cyst from the granuloma. The cyst continues to enlarge by epithelial proliferation in the lining and by hydrostatic pressure generated in the cyst lumen from the hyperosmolarity created by cellular breakdown and sloughing of cells into the lumen [5].

Therefore, the osmotic gradient favours transudation of fluid into the lumen, which maintains its hydrostatic pressure and causes further resorption of the surrounding bone. This cycle can be broken and reversed in most situations if the inflammatory focus is removed, (i.e. root canal therapy, or removal of tooth). However, if the tooth is removed, the apical lesion should be removed as well [5]. RCs are usually asymptomatic, [19] or until the cyst becomes large, at this point, the cysts may cause swelling. In most cases the cystic lesions are discovered when periapical radiographs of teeth with non-vital pulps are taken; or are detected incidentally on plain panoramic radiographs. At first the enlargement is bony hard but as the cyst increases in size, the covering bone becomes very thin, despite the subperiosteal bone deposition and the swelling then exhibits “egg-shell cracking” only when the cyst has completely eroded the bone will the lesion be flucuant. In the maxilla, there may be buccal or palatal enlargement, whereas in the mandible it is usually labial or buccal and only rarely lingual [1].

The occurrence of inflammatory cysts in mandibular first molars is due to high prevalence of caries. These teeth have deep pits and fissures on the occlusal surface, large-sized crown which leads to accumulation of acid produced by bacteria, and the early eruption of the tooth. Also Lack of oral hygiene and prompt treatment could influence the development of caries.

It is often said that RCs are painless unless infected. The overlying mucosa is initially normal, however, later on becomes discoloured with dark blue tinge [20]. They can cause mobility and displacement of teeth and once infected, lead to pain and swelling, after which the patient usually becomes aware of the problem [1,21].RCs on the lateral margin of a root in association with an accessory root canal must be differentiated from a lateral periodontal cyst [5]. Quite often, more than one RC may be found in a patient and this has led a number of authors to believe that there are cyst prone individuals who show a particular susceptibility to develop RCs [22].This view is supported by the fact that RCs are relatively rare in relation to the vast number of carious teeth with dead pulps. It is possible that an immune mechanism may inhibit cyst formation in most individuals and that cyst prone subjects have a defective immunological surveillance and suppression mechanism. It is also possible that some individuals have a genetic tendency to develop RCs. Multiple RCs may also be seen in patients with hereditary dental defects, but in these cases this is because of morphological defects resulting in early exposure and death of pulp [1].

The bilateral occurrence of odontogenic cysts is rare. It could be suggestive of developmental etiology. However, this is not widely accepted because inflammation of the cystic lining has been a consistent feature [23].Joshi et al. [24] reported an unusual case of bilateral RCs associated with mandibular right and left first molars in a thirteen years old child. The patient was subjected to surgical enucleation of cystic sac. Holla et al. [25] presented a case of sixteen years old male patient who had bilateral RCs symmetrically present in the mandible.

The classical description of radiological appearance of RCs is that they are round or ovoid radiolucencies of variable size which are well surrounded by a radio-opaque margin [1]. In infected or rapidly enlarging cysts, the radiopaque margin may not be present. Lesions are usually 1.0–1.5 cm in diameter and rarely exceed 3 cm. The walls vary from extremely thin to a thickness of above 5 mm. The wall is fibrous and will often contain numerous capillaries, particularly in areas adjacent to the epithelial lining [5].

Adjacent root resorption in RCs is usually absent, however, infected and long standing cysts may exerts pressure adjacent roots. Loss of vitality may be seen if the lesion is large involving adjacent teeth. Usually RCs are unilateral, bilaterally symmetrical involvement was noticed in few cases. This could be attributed to the initiation of cystic lesion at the same time.

The contents of RCs are usually, a soft brown material, often with glistening, oily, yellow flecks. Nodules of opaque yellow material, representing cholesterol, may be seen protruding into the lumen or within the wall. These cholesterol crystals are found in many RCs, but not in all RCs. These impart a shimmering gold or straw colour to the cystic fluid [20].

The cavity of RC is generally lined by stratified squamous epithelium of variable thickness usually showing spongiosis and hyperplasia. Microscopically, a RC is limited by a mature collagenous connective tissue wall. Abundant fibroblasts can be identified within the cystic wall. The inflammatory cell infiltrate in the wall in the proliferating epithelial linings is of variable degree and consists predominantly of polymorphonuclear leucocytes whereas the adjacent fibrous capsule is infiltrated mainly by chronic inflammatory cells [26,27].

Lymphocytes are generally the most prominent cells in the infiltrate and are characterized by their darkly stained nucleus, which occupies most of the cytoplasm. Plasma cells are also abundant in cyst wall and mostly seen in long standing cysts. These are characterized by an eccentric nucleus with a chart-wheel arrangement of the nuclear chromatin, particularly in the subepithelial zone. Other histological findings within the cystic wall are: erythrocytes and areas of he-
morrhage, occasional spicules of dystrophic bone, multinucleated giant cells and cholesterol crystals.

Browne [28] postulated that the main source of cholesterol was from disintegrating red blood cells. Cholesterol accumulates in the tissues because of relative inaccessibility of normal lymphatic drainage. Arwill and Hayden [29] confirmed the origin from red blood cells. They showed that the crystals may form in congested capillaries in the inflamed areas as they appear to be enveloped by endothelial cells.

In histological sections, the cholesterol crystals are dissolved out and clefts are seen surrounded by dense aggregations of multinucleate giant cells. The granulation tissue containing the cholesterol protrudes into the cyst cavity and appears macroscopically and microscopically as a ‘mural nodule. Once the entire mass has passed into the cavity the epithelial breach heals and the cholesterol crystals lie free in the cyst fluid. An interesting and peculiar structure, originally described by Rushton and subsequent reported by Shear and others, is the hyaline body or Rushton body [30].

These are the hyaline structures that are eosinophilic and brittle in consistency. They may be linear, curved, hairpin, or round shaped. These hyaline bodies are tiny linear or arch shaped bodies, generally associated with the lining of the epithelium [5]. Russel bodies (round eosinophilic globules) can be seen within or outside the plasma cells and are the result of very active immunoglobulin synthesis [5].

Rarely RCs may be lined by mucous producing epithelium in either maxillary or mandibular locations. This is the result of metaplastic transformation of the epithelial rests of Malassez which are pluripotent. Remnant of odontogenic epithelium may be found in the fibrous capsule and there have been reports of examples where epithelial proliferation is so extensive that it resembles squamous odontogenic tumour [31]. In rare instances, as reviewed by Gardner a few well documented cases have been reported which indicate that squamous carcinoma may occasionally develop from the epithelial lining of radicular and other odontogenic cysts [32]. Several studies have indicated that it is not possible to rely on the radiographic size of a periapical radiolucency to establish the diagnosis of either a cyst or a granuloma unless the lesion is larger than 2 cm in diameter [33]. Rarely, RCs induce resorption of the root of the affected tooth. The most important aspect for the diagnosis of RC is related presence of tooth with non-vital pulp [1] with periapical radiolucency [5]. Periapical radiographs and pulp tests are the most reliable diagnostic aids. The treatment of RCs is dictated by mostly the restorability of the tooth. Treatment of non-restorable teeth involves removal of the involved tooth and careful surgical curettage of the periapical area. Restorable teeth are treated with endodontic therapy followed by full occlusal coverage restorations [5]. Under some conditions root canal therapy may be carried out with apicectomy of the associated tooth and excision of the cystic lesion.

The cyst does not recur if surgical removal is adequate. If the cystic sac is badly fragmented, leaving epithelial remnants, or if a periapical granuloma is incompletely removed with epithelial rests remaining, a residual cyst may develop after months or even years later. If untreated, the RC slowly increases in size at the expense of the surrounding bone. The bone undergoes resorption, but seldom is there a remarkable expansion of the cortical plates, as is frequently seen in the case of the dentigerous cyst. RCs definitely resolve if the tooth and the apical lesion are removed. If the tooth is removed and the cyst is not, most cysts will involute because of the removal of the inflammatory focus. A few rare cases will retain their cystic stimulation independent of the tooth, probably by ongoing inflammation in the wall of the cyst. This is termed a “residual cyst” [5].

4. Conclusion

The RCs are the most common inflammatory odontogenic cysts and are commonly found in the maxillary anterior region. Small cysts are usually found associated with non-vital teeth which are frequently involved with trauma. The RCs in mandible “usually go unnoticed and rarely exceed the palpable dimensions and are detected on routine radiography. Quite often more than one RCs may found. Treatment of choice is endodontic treatment or extraction of the offending tooth and excision of the cyst. The present case of a fourteen years old female illustrates large RCs in the mandible with bilateral symmetrical occurrence treated by excision and extraction of them involved teeth successfully.

References


