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Case report

ENUCLEATION OF A GIANT RESIDUAL CYST WITH INFERIOR ALVEOLAR INVOLVEMENT: CASE REPORT WITH A 2-YEAR FOLLOW-UP

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ABSTRACT

Residual cysts are inflammatory cysts related to dental extractions in the maxillae. Most of these lesions involve edentulous areas affected by previous surgical extractions where inflammatory cysts already existed. Residual cyst presents as well-defined radiolucency with clear margins without erosion of neighboring structures. Rhizolysis, defined as the process of resorption of hard tissues of the dental element, appears to be physiologic during permutation, pathologic in permanent elements, and a possible consequence of many dental treatments. This clinical report aims to describe a surgical treatment of a large cyst resulting from a previous extraction using a simple clot as a cavity filler. Based on clinical, radiographic, and histopathological findings, the present case was diagnosed as an infected residual cyst. The lesion was surgically enucleated; preservation of all other teeth and vital structures was achieved, with no postoperative complications and satisfactory healing.

KEYWORDS: *cyst, maxilla, jaw bones*

INTRODUCTION

The maxillary bones are among the most affected by cysts because numerous epithelial remnants remain in close contact with the development of the dentition (1). Cysts can often have clinicopathologic and radiographic similarity, but they often differ in etiology and invasiveness, converging more toward what are termed neoformations (1).

Based on their origin, we classify them into odontogenic and nonodontogenic cysts. Odontogenic ones are the most common and develop from the epithelium of the early dentition. The epithelium of these cysts can be derived from the dental organ, Malassez remnants, reduced dental epithelium, and fragments of dental lamina. It is often the case that epithelial remnants can lead to the development of a residual cyst after removal of the affected tooth (2).

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Underlying the etiology of residual cyst is almost always a root cyst formed apically or adjacent to an extracted tooth (2). Therefore, they present the same clinic as inflammatory but differ histologically because usually the inflammatory infiltrate decreases and noninflammatory fibrous collagenous tissue is present in their walls (3). They also have a thin epithelial lining that makes their identification by histopathological methods more difficult. In the literature, many studies have reported that residual cysts show active growth patterns in areas that have been edentulous for several years, often silently without giving alarm. The variable behavior of these cysts highlights the importance of further detailed studies of these often-overlooked lesions (4).

It has also been reported that cysts rarely transform into squamous cell carcinoma, and even less is known about the success of different surgical methods in their management (5). The present clinical case presents the surgical enucleation of a residual cyst in an adult subject, with a 2-year follow-up.

CASE REPORT

A 47-year-old male presented to the author's center with a localized swelling of the right lower jaw (Fig. 1). The swelling, according to the patient, was insidious onset, gradually progressive, non-fistulizing, involving the buccal cortical, painful on palpation. The area was already missing element 4.6, previously endodontically treated 20 years ago, with external root resorption and obvious discoloration; external examination revealed diffuse swelling on the left side of the maxilla. The overlying skin was normal and painless, and there was no associated lymphadenopathy (4). Intraoral examination revealed an intact permanent dentition, swelling in the right lower alveolus. On palpation, the surface was smooth without obvious fluctuations and showed mild bony crepitus both buccally and palatally (6).

Fig. 1. *Intraoral photo of the buccal side with detail of the buccal swelling.*

Radiological examinations

An orthopantomography (OPG) and computed tomography (CT) scan were acquired (Fig. 2). Coronal and axial sections and three-dimensional reconstructions were performed. On OPG, the lesion appeared to have a rounded radiolucency with well-defined margins with mesiodistal extension and adjacency to the mesial roots of element 4.7 and the root of element 4.5, but without signs of rhizolysis.

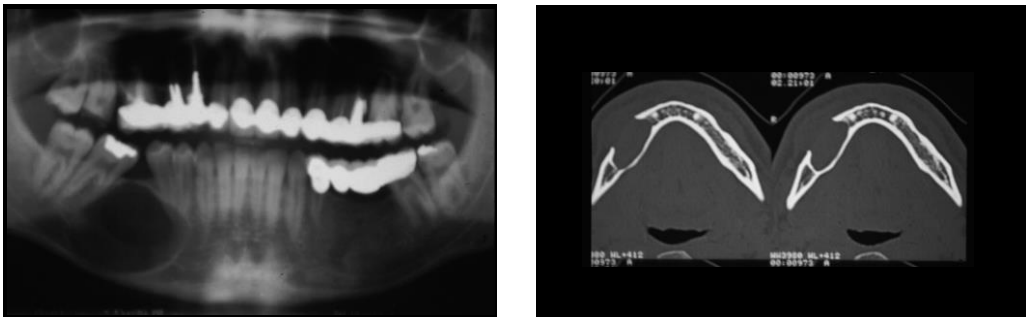

Fig. 2. *OPG and CBCT demonstrate the presence and extent of the cyst.*

CT scan revealed an expansile, primarily cystic lesion measuring 13.5 mm x 12.8 mm x 11.6 mm in maximum size that had led to thinning and erosion of bone along the cyst walls in several areas. Laterally the lesion was well circumscribed. The patient's biochemical parameters and investigations were all within normal limits (6). Ancillary tests revealed normal levels of serum calcium, phosphorus and alkaline phosphatase (7-9). Based on the clinical and radiological picture, a provisional diagnosis of residual cyst was made. In view of its clinical characteristics, which are similar to some lesions commonly found in the oral cavity, the differential diagnosis of radical cysts should include dentigerous cyst, Pindborg's tumor, periapical cementoma, traumatic bone cyst, ameloblastoma, odontogenic keratocyst and odontogenic fibroma. The confirmatory diagnosis of a residual cyst is established only after surgical biopsy and histopathological examination of the lesion (8,10-12).

Management and treatment

Since the involved bone had multiple cortical erosion involving the buccal wall, the enucleation of the lesion was planned with tooth extraction #4.5 (Fig. 3). After lifting a thick mucoperiostic flap, the margins of the cystic lesion were revealed.

Fig. 3. *Full-thickness (osteomucosal) detachment of the flap with evidence of the buccal cortical bone structure and the wall of the cystic lesion.*

The lesion was completely removed. The defect was filled with the simple native clot without the use of biomaterials. The area was completely sutured with non-reabsorbable sutures. Appropriate antibiotics (Amoxicillin 875 mg+clavuranic acid 125 mg), pain medications (Naproxen 550 mg tablet), and dietary instructions were provided.

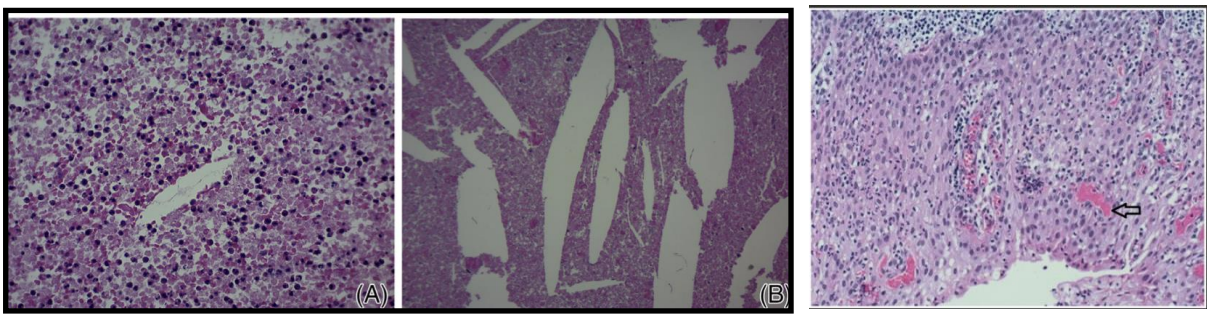

Fig. 4. *Cyto-histological analysis: arcading pattern of epithelium and chronic inflammatory cells with the presence of cholesterol cleft and erythrocytes.*

The surgical wound healed well after 20 days (13-15). After surgical enucleation and biopsy, histopathological examination revealed a non-keratinized layered squamous epithelium with long irregular-net-like ridges showing a characteristic arc pattern (Fig. 4). The underlying connective tissue was vaguely fibrocellular with a chronic inflammatory infiltrate containing predominantly lymphocytes and plasma cells. Many newly formed blood vessels with areas of bleeding have been observed. Histological results confirmed the clinical diagnosis of the root cyst. The success of surgery and the total recovery of bone volumes are highlighted in Fig. 5 at the 2-year follow-up.

Fig. 5. *Intraoral photo and OPG demonstrating the perfect healing of the soft tissues and the continuous ossification process of the former cystic cavity at 2 years follow-up.*

DISCUSSION

Residual cysts often have no symptomatic relevance and for this reason only a few detailed case series are available in the literature (16). The frequency of residual cysts has been reported to range from 1.4% to 18% of odontogenic cysts in the jaws. With the present clinical case, the damage resulting from the presence of cysts was evaluated. It is necessary to measure the size of the preoperative defect and the amount of defect after enucleation because the area, size and contour of the residual bony cavity influence the type of flap and any releasing incisions (17). The pathogenesis of radicular and residual cysts is similar, because the process begins with the spread of bacteria from a non-vital tooth into the periapical region of the jaw bones. If left untreated, this infection leads to the formation of a periapical granuloma that contains activated T cells that produce cytokines. These cytokines act on epithelial remnants leading to proliferation of these remnants and differentiation into cyst formation. The proliferating epithelium becomes edematous through the accumulation of fluid and coalesces to form microcysts lined by epithelial cells with inflammatory infiltrate (18). The wall of the cyst has a semipermeable membrane and therefore the cyst increases in size by osmosis. Furthermore, lytic products of epithelial and inflammatory cells increase the osmotic pressure within the cyst leading to further expansion and the formation of large intraosseous cysts (19).

Residual cysts occur mainly in middle-aged patients in the third decade of life. Most authors report that the lesion has a slightly greater predilection for males, as shown in this case report. A review by Kambalimath et al. showed in their sample that males are 3.5 times more affected than females (9).

Clinically they are often asymptomatic and can be detected incidentally on routine radiographs, such as an OPG or intraoral x-ray. However, if the cyst becomes secondarily infected, patients may report pain and swelling and become aware of the lesion. As the cyst gradually increases in size, it can cause shifting and mobility of the teeth (20-22). The patient treated in this case report comes in for pain and swelling, but the symptoms are not specific as other odontogenic lesions may have a similar presentation (23).

The culprits are therefore bacteria, their toxins, and the products of bacterial metabolism, which can reach the periapical tissues (periodontal ligament and alveolar bone) through the internal canals of the tooth, previously affected by chronic infectious pathology (pulpitis), or by necrosis, in turn almost always caused by deep caries, more rarely following trauma or deep non-carious lesions (24-25).

One of the most common complications is empyema or cystic cavity infection. In this case the typical manifestations linked to an acute infection will appear, difficult to distinguish from those of a simple alveolar abscess, with pain, swelling and increase in body temperature. Fistulization can occur in the oral cavity or, more rarely, in the skin of the face. In cases of large cysts, massive growth can lead to compression of local nervous structures, resulting in the

appearance of paresthesias and subsequent disappearance of sensitivity, and to the deformation of bone and facial structures, due to its progressive externalization. Sometimes the cystic lesion can reach such a size that it poses the risk of fracture of the affected bone. Development of ameloblastoma and squamous cell carcinoma from a radicular cyst has been reported (26-29).

In the case of very large lesions, it is necessary to proceed with surgical enucleation of the entire lesion (cystectomy or Partsch II), or, in cases in which this is not advisable due to the risk of damage to vascular-nervous bundles. The opening in the oral cavity and marsupialization (cystotomy or Partisch I) are performed (30).

The first type of operation (cystectomy) involves exposure to bone breach (31-33) and the total removal of the cystic tissue. Therefore, the closure of the cavity may involve the use of a filler (autologous or heterologous bone) to facilitate healing (31, 34, 35). The advantage of this procedure is the timing and the possibility of histological diagnosis of the entire lesion, factors that make it the first surgical choice therapy in all cases in which there is no risk of injury to the vascular and nervous bundles. In all cases, the primary cause (i.e., the infection of dental origin) must be eliminated with adequate endodontic treatment or extractions (36, 37). The recurrence rate is 3-5% within a short postoperative period.

Guided bone generation methods are in use to assist the repair process after surgical enucleation. However, there is considerable controversy regarding the use of guided bone regeneration techniques in periapical defects (38). Few studies are of the opinion that regenerative techniques are not superior, both in speed and quality of healing (39). On the contrary, other studies (40) have stated that the results of conventional treatment were lower than expected compared to cases in which regeneration methods were used (28). Among all odontogenic cysts, radicular/residual cysts have the highest potential for malignant transformation of their epithelium. It was reported that residual cysts were the cysts most frequently transformed (60%) into squamous cell carcinoma, followed by dentigerous cysts (16%) and odontogenic keratocysts (14%) (6). This further highlights the importance of performing a biopsy.

The pathogenesis of malignant transformation of odontogenic cystic epithelium remains unknown (6). Some authors have proposed that prolonged chronic inflammation may be a precursor factor for malignant transformations in the cyst epithelium. This finding is supported by the presence of chronic infiltrate of lymphocytes and plasma cells in the cystic lining of malignant cystic epithelium (41).

CONCLUSIONS

Residual cysts present with clinical-pathological characteristics similar to more aggressive cystic lesions. Their timely management is of extraordinary importance. Biopsy and histopathological examination are the main methods of accurate diagnosis, along with radiological correlation.

Enucleation is an appropriate surgical method for the management of residual cysts with a very low recurrence rate without the need for bone regeneration techniques. In this clinical case, it was possible to obtain excellent healing of the bone structure and tissues at 24 months without the use of more invasive regeneration techniques.

Consent

Written informed consent was obtained from the patient for publication of his clinical details and clinical images.

Conflicts of Interest

The authors declare no conflicts of interest.

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