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Research Article

Large Asymptomatic Maxillary Radicular Cysts Simulating Odontogenic Keratocysts or Bone Tumor Lesions: Importance of Radiographic Investigation

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Abstract

Background: Radicular cysts are common odontogenic entities, accounting for about 50% of all odontogenic cysts. They are associated with non-vital dental elements in which deep caries or inadequate endodontic treatments allow bacteria to migrate and infect peri-apical areas. Subsequent phlogistic processes trigger embryological epithelial rests potentially present in periodontal ligament, making them proliferate and form a cystic lesion.

Materials and Methods: We reported 2 cases of patients with maxillary radicular cysts associated with non-vital elements. Both patients showed up for routine visit and had no symptoms related to their lesions. Only intra-oral examination and especially radiological investigation could assess the presence of the lesions, allowing further histological investigation to reach a final diagnosis. Treatment included a surgical approach in both patients: cyst was removed and affected teeth or radicular residues were extracted.

Results: A few months later, a post-surgery OPG x-ray was performed and the bone structure appeared completely regenerated as no osteolytic lesions could be detected.

Conclusion: Radicular cysts often affect patients that aren't aware of their presence, as clinically these entities show no symptoms until very late stages or if infective complications arise. An accurate intra-oral examination and more importantly routinary basic radiographic imaging, e.g., OPG, can identify early maxillary lesions and guide clinicians in their diagnostic approach.

Keywords: Asymptomatic; Maxillary Radicular; Odontogenic; Radiographic

Introduction

Maxillary cysts are benign lesions consisting in pathologic cavities filled with fluid or semi-fluid substance. Cysts are surrounded by epithelium which is supported by connective tissue. Given their different etiology, cysts have undergone various classifications over time, being the most recent the "World Health Organization classification of Head and Neck Tumours 2017", which classifies them as follows (Table1) [1].

Among these lesions, the odontogenic cysts (both developmental and inflammatory) are more frequently detected in clinical practice [2]. They present topographic and histological relationships with the dental element involved and its residual developmental tissues, which are triggered by stimuli or degenerative processes. The embryological epithelial residues at the origin of cystic lesions are

Odontogenic cysts of inflammatory origin
Radicular cyst (RC) Inflammatory collateral cysts (ICC)
Odontogenic and no odontogenic developmental cysts
Dentigerous cyst (DC) Odontogenic Keratocyst (OKC) Lateral Periodontal cyst (LPC) and botryoid Odontogenic cyst (BOC) Gingival cyst (GC) Calcifying Odontogenic cyst (COC) Orth keratinized Odontogenic cyst (OOC) Nasopalatine duct cyst (NDC)
Bone cyst
Aneurysmal bone cyst (ABC) Simple bone cyst (SBC)

Table 1: World Health Organization classification of Head and Neck Tumours 2017.

- The Serres glands that originate from dental lamina,
- Post-functional ameloblasts and reduced enamel epithelium,
- The Malassez residues that survive the fragmentation of the Hertwig epithelial sheath [3].

The literature suggests 2 theories regarding the pathogenesis of inflammatory cysts, being the "hydrostatic theory" the most valid: a degenerative stimulus induces epithelial residues that line the initial cavity to proliferate and secrete, allowing proteins, cholesterol residues and cells to accumulate while increasing cavity's osmotic pressure; an increased concentration of solutes causes the passage of liquids through the cystic wall inside the cavity; the passage of liquids through the cystic wall inside the cavity is induced by the increasing of the dissolvable substances; in the end, the osmotic pressure activates osteoclasts, allowing the cavity to expand at the expense of the bone structure [4]. Along with this mechanism, the "prostaglandinic theory" is not unfounded, as prostaglandins generated from the cystic wall are likely to be involved in osteoclastic activation as well [5].

The final diagnosis for maxillary cysts is histological. Depending on the type of cyst, its clinical behavior may be different and it's characterized by different severity in the local expansion. Radicular cysts, which tend to grow slowly, are often asymptomatic and their presence is usually detected as an incidental radiography finding. If the lesion expands to the point where signs of inflammation appear, a surgical treatment is required: it could include the enucleation of the cyst, its marsupialization or a combination of the 2 techniques [5].

Radicular cysts are classified as inflammatory odontogenic cysts and they are overall the most common lesions of this type [6]. They arise from epithelial cell rests of Malassez of periodontal ligament that are induced to proliferate by inflammatory mechanisms, such as necrosis of dental pulp or a previous inappropriate root canal treatment: both events lead bacteria to invade root apices and cause the release of mediators which stimulate epithelial remnants, if present.

In this article we present 2 cases of maxillary radicular cysts and their treatment. This study was conducted in accordance with the principles of the Declaration of Helsinki and informed consents were obtained from both patients prior to enrollement.

Case Presentation

A 78-years old asymptomatic female patient came to our attention. During an intraoral examination, a tumefaction was identified apically to elements 1.5 and 1.6 (Figure 1), covered by mucosa of normal color. Both dental elements presented negative vitality, 1.5 was restored with a full dental crown, only residual roots of element 1.6 were present.

An OPG X-ray was prescribed and it revealed a rounded, unilocular and radiolucent lesion with a well-defined margin (Figure 2). To identify the margins of the lesion a CBCT was performed and it showed the presence of a 20x25 mm osteolytic lesion involving the apexes of elements 1.5 and 1.6's residual roots'. The lesion had deteriorated the vestibular bone cortex, whereas it was determinated that the involvement of the maxillary sinus floor could only be assessed intraoperatively.

After a comprehensive anamnesis, the patient was informed about the possible treatments and accepted a surgical approach: the cyst was enucleated and the relevant tooth parts were extracted.

Figure 1: Intra-oral view showing a swelling apical to 1.5 and remnant roots of 1.6.

The day before the surgery the patient started antibiotic, cortisonic and anti-inflammatory therapies, which she continued for 7 days.

After applying local anesthesia with Mepivacaine and vc 2%, a trapezoidal flap incision was performed with discharges mesially to 1.3 and distally to 1.6. After performing a full thickness dissection to expose the cyst and drain its caseous fluid, the cys-

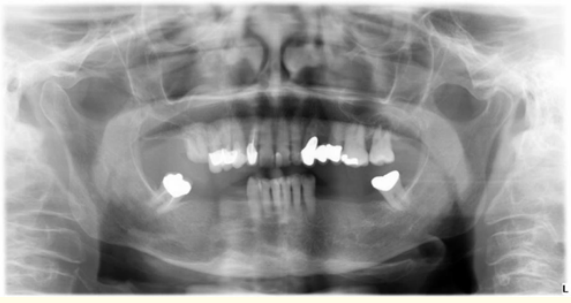

Figure 2: Initial ortopantomography revealing an osteolytic lesion apical to 1.5 and roots of 1.6.

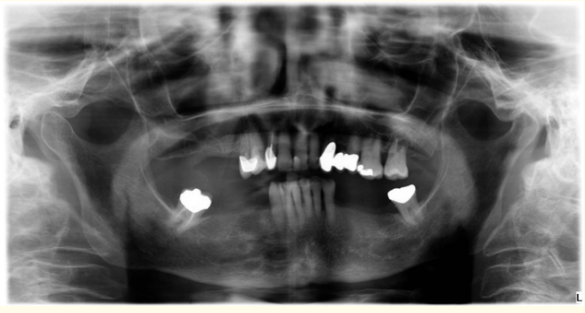

Figure 4: Ortopantomography 4 months after surgery showing complete bone regeneration.

tic membrane was accurately removed. Curettage was performed and the cavity was cleansed with an antibiotic solution and sterile physiological solution (Figure 3). Finally, to realize hemostasis and achieve a healing by primary intention, 4/0 Vicryl suture and compression were applied.

Figure 3: Intra-operative view showing the cavity that contained the cyst.

The patient was discharged with a stable clot. The histological sample was sent to the pathological anatomy laboratory of the San Paolo Hospital, suspecting a diagnosis of keratocyst.

A 7-days follow up visit was given to the patient, as well as post-surgery oral hygiene instructions.

For a follow-up evaluation an orthopantomography was requested 4 months after surgery and it showed spontaneous bone regeneration after the removal of the lesion (Figure 4).

Case Presentation

A 66-years old male patient requesting a full-mouth rehabilitation came to our attention. Patient's medical history included diabetes. Many dental elements were missing, those present had severe mobility. Patient was asymptomatic, without signs of tumefaction. During intraoral exploration it was possible to assess the absence of the maxillary external cortical bone in the second quadrant. An OPG x-ray and subsequently a CBCT (Figure 5 and 6) were requested, revealing a well-defined rounded osteolytic lesion with a major axis of 3,5 cm, extending in the maxilla and apical to 22, 23 and 24 elements. 22 and 24 were both necrotic, while 23 had previously undergone endodontic treatment. Element 23 was negative on the percussion test, and the restoration did not appear to be infiltrated by caries. This led us to exclude a possible odontogenic etiology of the radiolucent lesion starting from element 23.

It was scheduled an oral hygiene session before the surgery; antibiotic and anti-inflammatory therapy was prescribed to the patient.

On the day of surgery, under local anesthesia a flap was created for the extraction of teeth 12,11,21,22 (Figure 6); subsequently an osteotomy and enucleation of the cyst were performed (Figure 7), while the sample was sent to the laboratory for further investigations. A GBR was performed with biomaterial and membrane, Vicryl sutures were applied to achieve healing by primary intention (Figure 8).

At the 2 weeks follow-up appointment the stitches were removed.

On that date it was also decided to extract 1.3 and 2.3 and to take an impression in order to start the rehabilitation with an upper prosthesis; a photodynamic and glycin therapy was also prescribed for the patient's peri-oral burning symptoms; clobesol and hydroxymethylcellulose-based galenic preparation was recommended for oral application.

In the meantime, the patient had moved, making it impossible to record a regular post-operative RX.

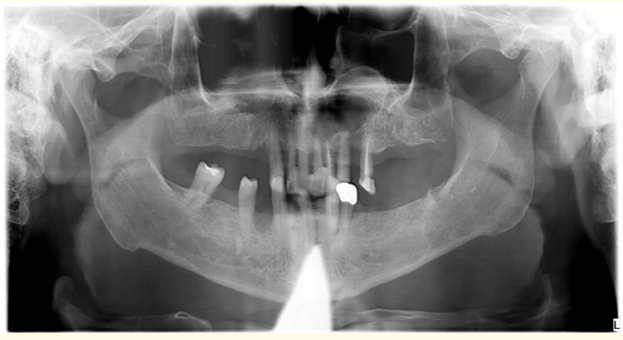

Figure 5: Initial ortopantomography revealed a large osteolityc lesion apical to 2.1, 2.2, 2.3 and 2.4.

Figure 6: Axial scan, TC panorex, DVT image and oblique cross-sections obtained from CBCT

Figure 7: Intra-oral view after performing cyst removal and cleansing of the cavity.

Figure 8: Intra-oral view of the surgical site with application of Vicryl sutures.

Discussion

Although maxillary cysts are considered uncommon in daily practice, these lesions constitute a large percentage of samples in oral and maxillofacial pathology practice, as Tamiolakis., *et al*. (2019) stated in their 38-years retrospective study about the demographic and clinical characteristics of 5294 jaw cysts in a single Oral Pathology Department [2].

The most common type of odontogenic cyst is the radicular cyst, which has been described in various studies as the most frequently diagnosed lesion [2,6]. This type of scenario is confirmed by the first epidemiological study on NJCs (non-neoplastic jaw cyst) in the Italian population according to 2017 WHO classification, where they represent the 56.6%, followed by dentigerous cyst (23.4%) and odontogenic keratocyst (12.9%) [7].

Radicular cysts are always associated with non-vital dental elements and develop when bacteria and their toxins invade the periapical area as a result of caries or previous inadequate endodontic treatments. An inflammatory process occurs, releasing mediators that stimulate the proliferation of epithelial cell rests of Malassez potentially present in the periodontal ligament, thus forming the cyst. Histologically, radicular cysts are covered with a nonkeratinized stratified squamous epithelium; the epithelium may vary in thickness from only few cells to a thick layer with significant hyperplasia [8].

It has been reported that radicular cysts more frequently develop in the maxilla and most cases involve the anterior teeth, es-

pecially the maxillary lateral incisor [9]. Their size ranges from 5 to 15 mm, although in the maxilla a cyst may expand for more than 15mm [10]; this evidence is confirmed in our 2 cases, where both cysts had respectively major axis of 2,5 cm and 3,5 cm.

Radiographically, a radicular cyst appears as a well-defined unilocular radiolucency often surrounded by a sclerotic border and it is closely associated with the apex of the affected tooth [11]. However, such radiographic features make it difficult to distinguish them from other osteolytic lesions that affect the jaws, be it other types of cysts or granulomas affecting teeth undergone endodontic failures [12] or bone tumours.

Differential diagnoses of radicular cysts are the following: odontogenic keratocyst, dentigerous cyst, traumatic bone cyst, aneurismatic cyst, ameloblastoma, odontogenic fibroma and central giant cell granuloma [13,14].

These entities share some radiographic features (radiolucency, smooth and sclerotic border) that cannot lead us to a definitive diagnosis but they should be considered in making the differential diagnosis.

Among the aforementioned lesions, an important entity to be put in differential diagnosis with the radicular cyst is the odontogenic keratocyst (OKC). OKC is the third most common cystic lesion and it accounts for about 7-20% of cystic lesions of the jaws [2]; it's been classified once again as a cystic lesion in 2017 by the WHO classification [1], although in a previous WHO classification (2005) it had been defined an odontogenic keratocystic tumor because of its local aggressive behaviour and high recurrence. This type of lesion, like ameloblastoma, is most often found in the posterior region of the mandible [15], although it can be found in the posterior maxilla [16] and more rarely also in the anterior region of the maxilla [17,18]. One study reports a single case of keratocyst originated from the temporomandibular joint [15].

In a study by Kitisubkanchana., *et al*. [19] radiological characteristics of OKC are outlined in comparison to those of ameloblastoma in differential diagnosis: radiolucent lesion, unilocular shape (82%), smooth borders, possible causing displacement of adjacent teeth (33.7%) and rarely root resorption (7%), limited bone expansion.

To note that the presence of multiple keratocysts, accompanied or not by other clinical manifestations, especially cutaneous, must raise the diagnostic suspicion of nevoid basal cell carcinoma syndrome (NBCCS), also known as Gorlin-Goltz syndrome [20].

As it occurred in our 2 cases, radicular cysts are often asymptomatic, being discovered by chance in basic radiographs such as orthopantomography. Hence it is crucial to prescribe periodically such basic and yet mandatory imaging modality, always in compliance with the most up-to-date guidelines on radiation protection. In fact, after performing anamnesis and an accurate clinical examination, only first level x-ray investigations enable clinicians to detect possible lesions of jaws' district [21], whereas second level radiological investigations (CBCT) are required as they have a greater diagnostic accuracy, especially for lesions of the maxilla [22].

As for the treatment, any management strategy must evaluate the cystic lesion from both histological and radiological point of view. A 3D evaluation of the cyst using CT allows the best assessment of its margins and the relationships to the anatomic structures [23]. However, the radiography itself does not differentiate cysts as it is not based on pathognomonic criteria; thus, the differential diagnosis is based on the histological analysis [24].

As seen in our 2 case reports, the histological analysis (Figure 9) was crucial in confirming the clinical and radiological suspects of odontogenic cyst of radicular type, showing its aspects of chronic diffuse intra- and peri-cystic phlogosis with lymphoplasmocellular and *histiocytic infiltrates associated with cholesterol residues.*

Figure 9: Enucleated cyst to be analyzed in the pathological anatomy laboratory of the San Paolo Hospital.

Treatment of radicular cysts implies their direct enucleation (Partsch II), marsupialization (Partsch I) or a combined technique [4].

Marsupialization is a surgical technique indicated in case of particularly large cysts. It consists of cutting the cyst and suturing its edges to oral epithelium to form an exposure of the lesion to the oral cavity; decompression and progressive volume reduction of the cyst follow, allowing a gradual growth of the surrounding bone tissue [25]. As described by Sorrentino., *et al*. in their article [26], in case of lesions of considerable size, direct surgical techniques require a more invasive approach and are susceptible to early and late complications such as iatrogenic mandibular fracture, risk for adiacent teeth to lose vitality, risk of damaging noble structures such as the inferior alveolar nerve. The success rate of marsupialization, as an initial step, in the treatment of very large cystic lesions allows a more conservative second surgical access [27]. Following exposure of the cystic cavity to the oral cavity, a histological change occurs in the epithelial lining of the cyst [28]. This, in fact, becomes thicker and takes on characteristics more similar to those of the normal oral mucosa, facilitating the subsequent enucleation of the cyst [26].

Endoscopically assisted enucleation is another innovative alternative method that can be as conservative as marsupialization, allowing preservation of important surrounding structures, with the greater advantage of one step treatment, reduced healing period and very low morbidity [13]. Endoscopic approach for maxillary and mandibular cysts provides a better vision, ensures complete cyst enucleation and reduces operative risks for adjacent vital anatomical structures [29].

To avoid recurrences, treatment also includes removal of the primary cause, i.e., the infected tooth or the infection it bears. So possible other treatments are endodontic treatments (or retreatments) with associated apicectomy, or extraction of the responsible dental elements if they are not treatable.

At the follow-up visit a radiographic examination (e.g., OPG) should be performed to assess the complete healing of the bone lesion. In fact, in addition to having a fundamental diagnostic purpose, radiology plays another primary role at follow up as it allows the examination of hard tissues undergone treatment and/or surgery. For example, a study reported that further radiographic investigation can give us information regarding the healing of the lesion in a patient who's then eligible for further treatment, such as *implant-prosthetic rehabilitation* [30]. In one study with patients who reported pain and swelling in the jaws and who were diagnosed with cystic lesions, radiographic imaging was performed before and after cystic enucleation to verify if there was spontaneous regeneration of bone tissue [31]. In another study, postoperative panoramic radiographs were prescribed to demonstrate that spontaneous bone regeneration could occur in large mandibular cysts without the aid of any filling material following enucleation [32].

Conclusion

Radiological diagnostic tools such as orthopantomography are able to intercept asymptomatic bone lesions very early. In the perspective of a medicine that is increasingly oriented towards prevention, radiological instrumental investigations are fundamental, to allow the clinician to intercept lesions, have an early diagnosis, implement timely therapy and so to have a better prognosis.

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