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Importance of Complete Clinical History in the Management of Parakeratinized Odontogenic Keratocyst: Case Report

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Abstract

Introduction: The Odontogenic Keratocyst considered potentially aggressive and of high recurrence located between 30 and 60%, with clinical and radiographic characteristics not clearly defined. It occurs at any stage of life. The 70 to 80% of the cases are located in the jaw, commonly in the area of the lower third molar and mandibular angle from where they progress towards the branch and the body. The lesions are long latent, often asymptomatic and can reach remarkable dimensions without significant deformation of the jaw bones. They are often found during the routine radiographic examination.

Objective: To report a case of Odontogenic Keratocyst, evaluating it's clinical, radiographic and histo pathological characteristics that lead to conservative management and treatment.

Case report: A 40-year-old woman presented with pain on contact in the region of the lower right side of the face and a sensation of leakage of bad-smelling fluid in the posterior and inferior region of the mouth. X-ray and tomographic examination revealed a large expansive lesion of both cortices, compromising the posterior region of the mandibular body, extending to the angle and lower half of the right mandibular branch, also compromising piece 4.7 in its distal and palatal regions. After the histo pathological examination a definitive diagnosis of parakeratinized Odontogenic keratocyst was reached. Surgical surgical treatment of complete enucleation of the lesion was obtained, obtaining satisfactory results.

Conclusion: The meticulous elaboration of the clinical history based on clinical, radiographic and histopathological findings helps to arrive at a correct diagnosis, the same one that allows elaborating an adequate treatment plan.

Keywords: Parakeratinized Odontogenic Keratocyst; Odontogenic Tumor Keratocyst; Diagnosis

Introduction

The Odontogenic Keratocyst (OKC) was defined by the World Health Organization (WHO) in 2005 like a benign and intraosseous tumor of Odontogenic origin, with a characteristic lining of stratified Parakeratinized squamous epithelium and a potential of aggressive and infiltrative behavior [1,2]. This term was recently changed by WHO, being clearly considerate in the category of cyst [3]. The OKare an independent clinical entity with a typical microscopic pattern of aggressive clinical growth, biological behavior of high recurrence from 30% to 60%they arise from the proliferation of the epithelial dental lamina of the jaw and maxilla [2,4,5]. Never the less, it is suggested that they may also derive from an extension of the basal cell component of the buccal epithelium that covers them, as well as the dental organ by degeneration of the stellate reticule, before the enamel apposition begins [6,7].

The OKC is a fairly common clinical finding, accounting for 10-20% of Odontogenic cysts and is the third most common jaw cyst [4,8]. They appear in all ages, although the most accurate diagnosis is common between the second and third decades of life [2]. Their most frequent location in a 70-80% is the mandible, commonly at the level of the lower third molar and mandibular angle (50 to 75%) from where they progress towards the branch and mandibular body with higher percentages of occurrence in the male sex [9-11].

Clinically it is an asymptomatic lesion, of long evolution, slow and expansive growth not destructive, the skin and the mucosa present normal characteristics, crackling to the palpation. Frequently this injury is associated with impacted teeth [11]. When the lesion is advanced, there is an increase in volume that compromises a bone table (lingual in the mandible and vestibular in the upper jaw), being able to reach large dimensions since it grows more through the medullary spaces than transversely, accompanied by a mass and pain [2,9].

Radio graphically they present a variable appearance, being able to give an oval or rounded cavitary aspect, unilocular in its great majority and with less presence of multilocularity, mainly in large lesions and in more than 40% it is associated with the crown of a retained tooth [4,10]. It rarely presents as interradicular and periradicular radiolucent image, and when it happens the adjacent teeth keep their vitality [12]. The OKC shows a radiolucent image, with well-defined and thin edges, which may or may not have peripheral sclerotic halo [12]. In many situations, the literature shows radiographic similarity to dentigerous cysts, due to the retention of dental pieces, multilocular lesions similar to ameloblastomas, residual cysts, lateral periodontal cysts and even radicular cysts [2]. Most of these lesions are usually unique, but exceptionally there can be multiple known as Gorlin-Goltz syndrome or also called basal cell nevoid carcinoma syndrome [13].

Histologically, the OKC shows two more frequent variants. The first of Parakeratinized aspect, showing thin stratified squamous epithelium (8 to 10 layers), with a Parakeratinized and corrugated surface layer, as well as an interface of epithelium - flat connective tissue, that is, without dermoepithelial invaginations. The basal cells have polarized, hyper chromatic nuclei and arranged in a row. The second, the orthokeratinized variant, which is less common, is characterized by having a thin epithelial lining, with a granular layer and another thick layer of uncorrugatedorthokeratin, basal cells that are not very evident and lumen usually filled with keratin-compatible material. The epitelial detachment of the cystic capsule is usually observed [1,14].

It is suggested that the same lesion may exhibit parakeratotic areas and orthokeratotic areas [2,5,10]. According to the histological study, the literature reports different clinical behaviors between the two most frequent variants. The orthokeratinized variant appears to be a different lesion that shows growth potential and a lower recurrence rate. It can frequently be associated with a retained tooth, have a unilocular radiographic appearance and be located preferentially in the maxilla or anterior region of the jaw [15].

The treatment of the OKC is based on the simple enucleating of the lesion and subsequent curettage. The most important purpose is to decrease the high rates of recurrence of this entity, which can occur up to ten years after surgery. Treatment options include simple enucleating (not recommended with high recurrence rates, 17-56%). Adjuvant techniques such as Carnoy's solution followed by enucleating (absolute alcohol, chloroform, 98% acetic acid and ferric chloride) can be used or the cryotherapy, decreasing in that sense the recurrence rates (1-8.7%) [16,17]. Likewise, decompression techniques derived from marsupialization prior to surgery have been used in order to decrease the size of the cyst and intraluminal pressure, allowing on that way a less aggressive surgery and with lower recurrence rates. In aggressive cases, a mandibular resection can be performed in block [2,10,16,17].

For all the characteristics presented above, this article describes the clinical and histopathological management and the planning of the conservative surgical treatment of a large right mandibular Odontogenic Keratocyst, observed in a 40-years-old woman that involves a continuous tooth, and its respective postoperative follow-up.

Case Report

Female patient of 32 years of age, native of Cusco, married, accountant of occupation, goes to Center of Stomatological Radio diagnosis Cusco (CERES), referred of Dental Clinic, in September of the 2017, for presenting light pain to the contact of right hemicara and sensation of discharge of liquid with bad odor of second inferior molar of the right side, with four months of

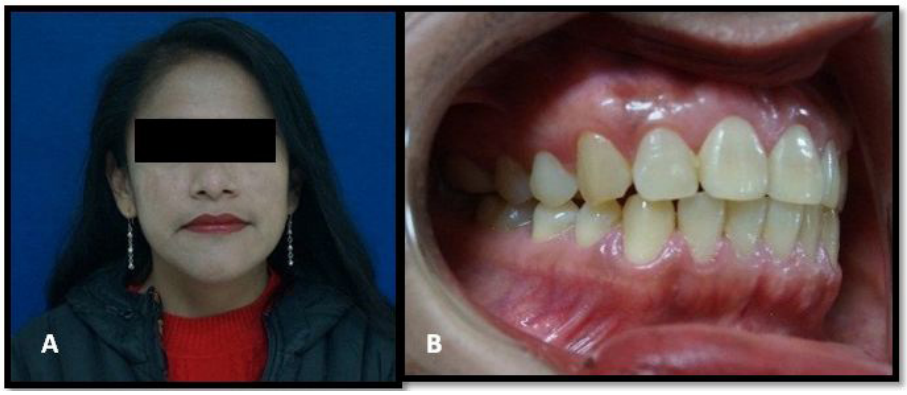

Figure 1A: Extra-oral view, showsslight facial asymmetry withVolume increase of right lower and posteriorhemicara,slight loss decrease on right nasolabial line
Figure 1B: Intra-oral view. Slight expansion of vestibular external corticalr, mucosa without color change, prominent

evolution approximately. Patient reports the dental extraction history of the third molar of the same región a bout a year ago, with out documentary evidence of radiographic diagnosis.

At the general clinical examination, slight facial asymmetry with increased right and lower right hemiferous volume was observed, with a slight decrease in right nasogenial line loss, painful on deep palpation and presence ofcrepitus, without signs of inflammation or palpable lymph nodes (Figure 1A).

At the intraoral examination, a slight increase in the volume of vestibular mucosa of the right lower molar region was observed without alterations in coloration but with the presence of prominent superficial blood vessels, mild external cortical expansion, crepitus and moderate pain on palpation, absence of a piece. 4.8, sensitivity increased to thermal tests of piece 4.7 with sensation of tingling to vertical percussion.A deep palpation of the region of the right internal mandibular ramus also showed greater crepitus and pain that radiated to the lower region of the latter (Figure 1B).

Panoramic radiography shows a wide lesion of approximately 5 × 4 cm in diameter, radiolucent, unilocular with defined limits, corticalized edges, projected from the distal root segment of piece 4.7, extending cephalad to approximately half of the mandibular ramus, engaging in its development piece 4.7and posterior region of mandibular body, right angle and right mandibular branch. There is an absence of part 4.8 and slight displacement of the mandibular canal.

In the Cone Beam Computed Tomography, the panoramic reformation can corroborate hypodense, extensive, defined edges and corticalized lesions that compromise the radicular segment of piece 4.7 (Figure 2A). The transversal reformation shows lesion of 22.01 × 10.28mm in size, oval shape; with cortical expansion and severe thinning of the lingual cortex with perforation of the latter in the inferior region (Figure 2B). In the sagittal reformation, complete exposition of the distal and palatal region of piece 4.7 with complete alveolar bone resorption of this region is evidenced, revealing in addition a slight displacement of the mandibular canal in caudal direction (Figure 2C).

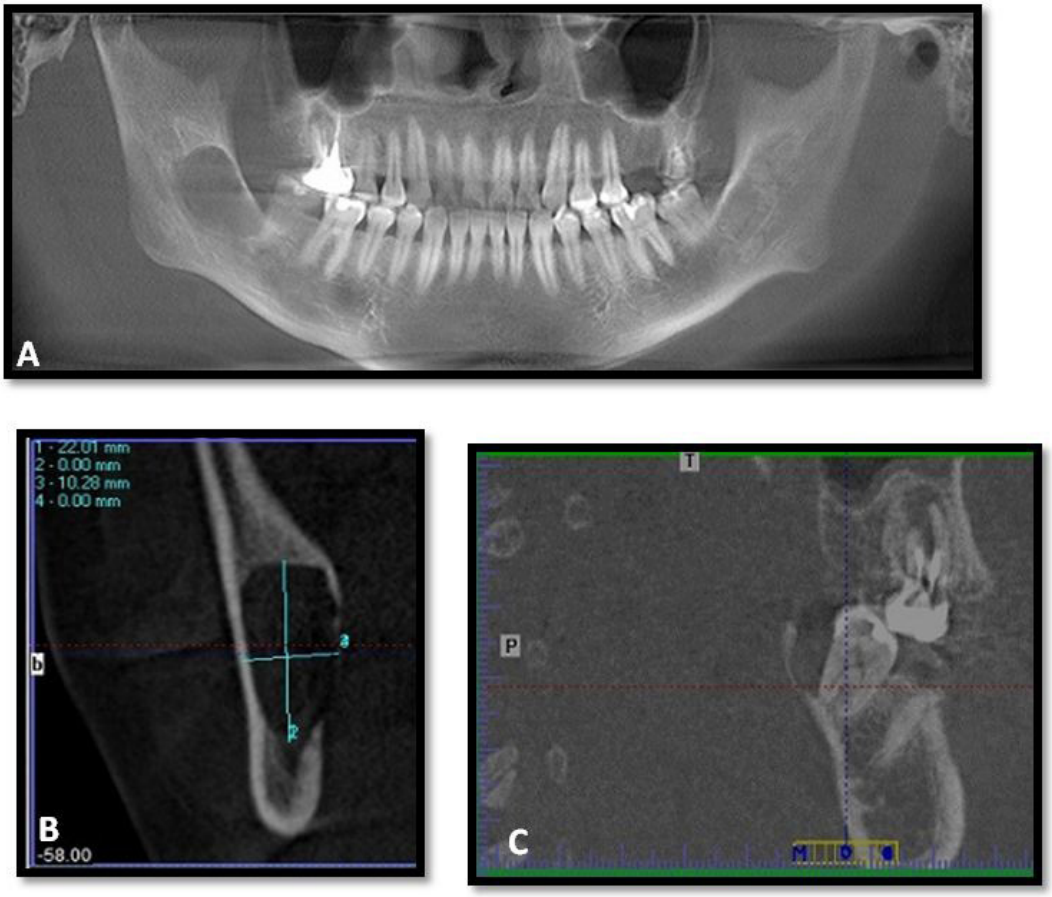

Figure 2A, B and C: Cone Beam Computed Tomography
Figure 2A: Panoramic reformation, hypodense lesion, extensive, with defined borders
Figure 2B: Transversal reformation, lesion size of 22.01 x 10.28mm, oval shape; cortical expansion, thinning and perforation of lingual cortical
Figure 2C: Sagittal reformation, exposure of distal and palatal regionsof piece 4.7

As a result of an exhaustive clinical and radiological examination, presumptive diagnoses of parakeratinized Odontogenic Keratocyst and ameloblastomawerereached, so it was decided to refer the patientto a specialized dental clinic for the irrespective treatment and follow-up.

Once the treatment plan was completed and the treatment plan was initiated, the patient's informed consent was signed and laboratory pre-operative examinations were subsequently requested, finding normal values, so it was decided to perform an excisional and aspiration biopsy.

Before performing the excisional biopsy, the aspiration of the lesion was planned, obtaining abundant bloody fluid and with remnants of tiny granules of apparent necrotic tissue, so we proceeded with the total enucleation of the lesion (Figure 3A). Under local inferior truncal anesthesia, a single retromolar incision was made to open the vestibular and lingual flaps, where the lesion was completely isolated and posterior enucleation, followed by careful curettage and washing of the bone cavity with abundant saline sterile solution (Figure 3B). To avoid recurrence, the extraction of piece 4.7 was performed and then sutured with polypropylene and treatment with analgesic, anti-inflammatory and intramuscular antibiotics and oral mouthwash without alcohol was indicated.

The withdrawn sample was placed in 10% formaldehyde solution to be sent for histopathological study (Figure 3A). The sections studied in the histopathological examination show flat stratified epithelium with inflamed stroma and thin surface of corrugated Parakeratinized area, with basal cells of hyperchromatic nuclei. In some areas the epithelium was separated from the cystic capsule and the lumen filled with keratin-like material, confirming the definitive diagnosis of parakeratinized odontogenic keratocyst.

Ten days after surgery, the first postoperative control and removal of the suture was performed. Three months later the patient was cited for clinical control and evaluation with panoramic radiography, showing excellent scarring and absence of tumor recurrence, showing bone neoformation in the region of the lesion (Figure 3C). Pulp vitality tests were performed on piece 4.6, which was positive.

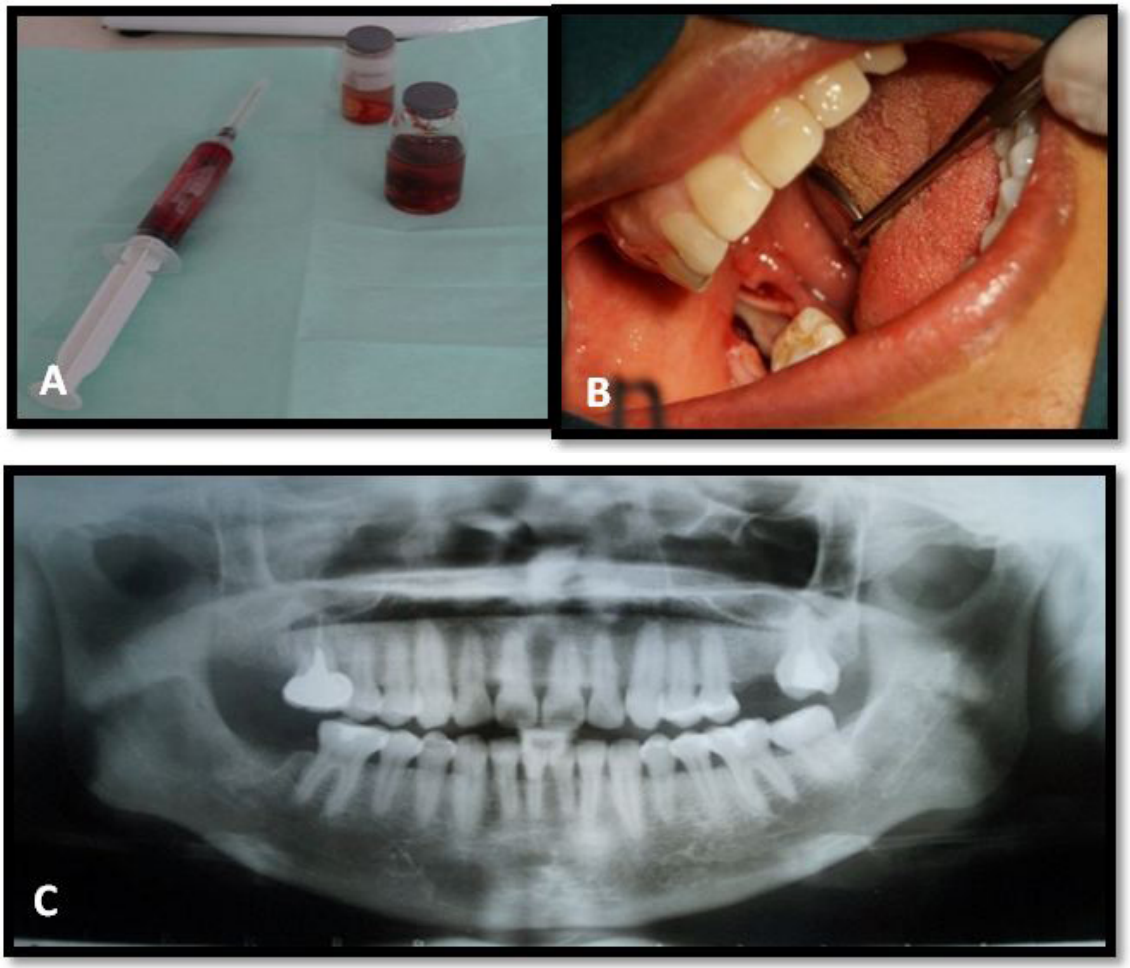

Figure 3A, B and C: Biopsy and treatment
Figure 3A: Excisional and aspiration in 10% formaldehyde solution
Figure 3B: Total enucleation of the injury
Figure 3C: Post-operative three months control

Discussion

The Odontogenickeratocystrepresents one of the most frequent odontogenic cystic lesion of growth, 10 to 15 % of those lesions approximately [2]. Depending on their histological origin, they can be presented in two forms: primordial origin (60% of cases)

derived from remains of the dental lamina, not associated with dental pieces and those of dentigerous origin (40%), which have their origin in the reduced organ of enamel and are associated with retained parts [16].

Most of the injuries reported in theliterature show prevalencebymale sex in a ratio of 1: 1.4; and its appearance related to any age, frequently diagnosed in the third decade of life and generally involves the posterior region of the jaw (50 to 75%), mainly of the third molars [1-3,10,16].Our case, on the other hand, reports on the appearance of an injury in a female patient and within the fourth decade of life, not being associated with a dental piece retained as it refers to primordial origin. In what does agree with the literature is its frequent location of presentation.

At the clinical examination the lesion can usually cause volume increase, pain and marked bone expansion, however, most cases are asymptomatic, a situation that does not agree with the one reported in this article, although there is no increase in volume marked, pain is observed at the contact as a greeting or superficial palpation, signs and symptoms that did not reflect the real state, but they can give an indication of the presence of pathology. Given this situation, this type of lesión presents clinical characteristics that mask these verity of the clinical picture, to which the literaturerefersto causal findings.

Radiographically, the multilocular appearance and defined and corticalized edges refer to the presentation of a parakeratinize dodontogenickeratocyst, while the unilocularaspectis more associated with the orthokeratinized type2, a definiti on that contradicts our report, becaus eitis of the parakeratinized type and unilocular, from this understanding weassumed that radiographically it would not fulfill defined parameters of presentation, the type of classification shown would be referred more from the histological point of view.

The treatments proposed in the literature are multiple, however the one that has presented lower percentages of recurrence, is the total enucleation of the lesion, followed by curettage, abundant washing with saline solution and exodontia of compromised parts, essential procedures performed in this article , which must have a postoperative follow-up of up to ten years to refer success and cure of the lesion and avoid recurrences that can be explained by many factors such as incomplete enucleation or curettage due to weakness of the cyst wall or invasion of soft tissues adjacent, or by the presence of satellite cysts given the high mitotic activity of this lesion that were not detected during surgery [2,3,10].

The reported case supports most of the clinical manifestations published in the literature, however we are facing a lesion that does not have defined characteristics in its entirety, which would make its recognition and accurate diagnosis, hence it is intended to highlight the importance of a history complete clinical, well elaborated, that allows to arrive at a correct and opportune diagnosis for its derivation to the specialist dentist and if it was necessary interconsultation with other medical specialties for its suitable treatment. Although most lesions usually have a benign presentation, they can generate large asymmetries and long-term facial deformities that would not only lead to esthetic problems but also psychological ones.

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