Case Report

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A Recurrence of a Giant Odontogenic Keratocyst: Radiographic Follow-Up

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Abstract

Odontogenic keratocyst could be a very aggressive lesion which may be imperceptible for the patient, not giving any type of symptomatology. This is usually discovered as a finding during a clinical evaluation of the maxillofacial area. The diagnosis of this lesion is merely histological, although it has some radiographic features that presumptively indicate the diagnosis. During a dental school brigade a patient came referring dental mobility, periodontal disease was absent and oral examination revealed a large swelling identified in the body of the mandible. Imaging studies revealed a large central lesion diagnosed as OKC when biopsy was performed. After enucleation and curettage of the lesion, radiographic follow-up was conducted every six months, at the third six month period recurrence of the lesion was present and surgical excision was performed with the recommendation of and strict radiographic follow-up in quarterly periods.

Keywords: odontogenic keratocyst, maxillofacial surgery, recurrence, histopathology, follow-up

Introduction

Odontogenic keratocyst is a lesion of developmental origin derived of the dental lamina or its remnants, or odontogenic basal cell hamartias. The given name was first introduced by Philipsen in 1956. OKCs have a predilection for males more than females, arising in the mandible more frequently than the maxilla with a pronounced peak frequency in the second and third decades of life [1]. Due to its neoplastic nature, terminology of this entity has been changed in the past, formerly called keratocystic odontogenic tumor the current term is odontogenic keratocyst as first was introduced [2]. The rate of recurrence for OKCs is of approximately 20-30% being more common during the first 5-7 years [3]. The presence of OKCs can be isolated

or syndromic as in Nevoid basal cell carcinoma syndrome (NBCCS) or Gorlin-Goltz syndrome where 75% of people with this condition present multiple OKCs [4]. Treatment for this pathology includes enucleation, marsupialization and curettage along with extraction of teeth where root resorption has developed, the use of Carnoy's solution and in some recurrent lesions enbloc resection may be required [5,6].

Case Presentation

During a dental school brigade a 32-year-old man presented with the concern of observing a gradual change in the position of some teeth for six months. During oral exploration dental mobility was identified without any sign of periodontal disease as well as a swelling under the

mucogingival junction in the vestibular area from right molar area to left premolar area of the mandible, patient referred no pain and he was unaware of this swelling. The patient was referred to the imaging department and orthopantomogram showed a large radiolucent lesion with extent from right first molar to left second premolar with mild root resorption affecting the teeth of the right side (Figure 1).

Figure 1: Initial Orthopantomogram.

The patient underwent incisional biopsy and diagnosis of OKC was established (Figure 2).

Figure 2: Histologic appearance of keratocystic odontogenic tumor. Stratified squamous epithelium with a parakeratinized surface.

Comprehensive evaluation of the patient was made and we begin preparing the patient for surgery. Cone Beam CT scan was performed with the purpose of determining

the vestibulo-lingual extent of the lesion and rule out perforation of the bony plates (Figure 3).

The patient was discharged from the hospital on the 2nd post operative day without any complication. Histopathology of the specimen revealed segments of dense fibrous connective tissue covered by parakeratinized stratified squamous epithelium with presence of satellite cysts in the capsule, thus confirming the diagnosis of OKC (Figures 5 and 6).

Figure 3: Axial bone CT scan shows expansion and thickening of body of the mandible.

The patient was operated under general anaesthesia and enucleation of the cyst was performed followed by a meticulous curettage of dental roots and walls of the surgical cavity. Using a surgical bur, adequate bone was removed from the bony cavity walls (Figure 4).

Figure 4: Enucleated cystic cavity.

Orthopantomogram evaluation was recommended every six months. The first six month evaluation showed osseous regeneration in about two thirds of the cavity (Figure 7).

Annual evaluation showed the same data although the remaining space to be filled with osseous tissue seemed to be more radiolucent than the first evaluation (Figure 8).

Figure 5: Histology of an Odontogenic Keratocyst.

Figure 6: Keratin Production by Satellite Cyst.

We decided to wait until the next evaluation to make conclusions and the patient was instructed to report any event such as swelling or increase in dental mobility. Third six month evaluation was conducted and orthopantomogram showed a well-demarcated radiolucent area in the apical right molar area extending to the incisors (Figure 9).

Figure 7: Orthopantomogram after 6 months of enucleation. Note significant bone formation in the floor of the cystic cavity.

Figure 8: Orthopantomogram after 12 months of enucleation, showing a diffuse radiolucency in the right molar area.

Figure 9: 18 months radiographic follow-up showing a well-demarcated radiolucency in the right molar area extending to the incisors.

Incisional biopsy was not performed since the patient referred swelling and discomfort in the area coinciding with the radiolucency. Outpatient surgical approach was performed to excise the recurrence of OKC followed by extraction of dental organs with root resorption. Enucleation of the cyst and peripheral ostectomy was performed. Histopathology of the specimen confirmed diagnosis of OKC and satellite cysts were identified (Figures 10 and 11).

Figure 10: Histopathology showing stratified squamous epithelium with a parakeratinized surface.

Figure 11: Satellite cyst containing keratin whorls.

Discussion

OKC highlight more than the other cystic lesions due to its aggressive clinical behaviour and its greater tendency to recur [7]. There are different predictive factors in relation to the recurrence behaviour of OKCs. Surgical techniques, patient age, location and size and shape are

clinical predictors, and histological predictors include parakeratinisation of the epithelial lining, subepithelial splitting and the presence of dental lamina remnants as well as satellite cysts. Further histological predictors include suprabasal splitting and basal layer budding [8]. In the present case parakeratinisation of the epithelial lining and the presence of satellite cysts was evident.

Various treatment modalities exist, preoperative imaging information and patient-specific decision contribute to determine the adequate approach [9]. Surgical treatment methods for OKC are categorized as conservative and aggressive. The conservative treatment includes enucleation, with or without curettage or marsupialization, preserving anatomical structures. Aggressive modalities include peripheral ostectomy, chemical curettage, or en bloc resection which is recommended for large lesions, syndromic patients and recurrences [10].

The present case confirms that OKC with specific histological features are more prone to recur and we emphasize that radiographic follow-up is essential since the recurrence of this entity is very unpredictable.

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