

Giant mandibular cyst : a case report

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ABSTRACT

Background: The odontogenic keratocyst (OKC) is currently known as a keratocystic odontogenic tumor by the World Health Organization (KCOT). It is described as a benign uni or multicystic intraosseous tumor of odontogenic origin with a distinctive lining of parakeratinized stratified squamous epithelium and the tendency for aggressive, infiltrative activity. This case report features a 54-year-old woman who has an unusual giant mandibular cyst that does not exhibit any of the standard characteristics of developmental odontogenic cysts.

Case management : The proper treatment in most cases is careful cyst enucleation and removal of the impacted tooth. In view of the large size of the lesion, treatment plan of enucleation was made by taking the cyst capsule in toto and can be treated with decompression

Conclusion: A case of Giant Mandibular Cyst in 54 female treated enucleation and removal of the impacted tooth is presented along with review of literature.

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INTRODUCTION

A cyst is described as a neoplasms in the form of pathological cavity filled with liquid, semi-liquid materials, or gas that is always covered by an epithelial layer and the outer part is lined by connective tissues and blood vessels.¹ The World Health Organization classifies neoplasms of the mandible as odontogenic cysts, nonodontogenic cysts, and pseudocysts.² A true cyst is characterized as an epithelial lined cavity, whereas pseudocysts lack an epithelial lining. The origin of the epithelial lining distinguishes odontogenic from nonodontogenic cysts. Odontogenic cyst epithelium develops from the tooth-forming organ. These include the decreased enamel epithelium, Malassez rests, and Serres rests.² Odontogenic cysts (OC) are further subdivided into developmental cysts, including odontogenic keratocysts (OKC) and dentigerous cysts, and inflammatory cysts, including radicular cysts.³

The most prevalent types of odontogenic cysts is radicular cysts (43,3%) followed by dentigerous cyst (25,5%) with relatively equal distribution for both sexes.⁴ In a separate investigation by Franklin et, al., the dentigerous cysts and odontogenic keratocysts (OKC) are the most frequent group of developmental cysts with the percentage of 27,5% and 14,2%, respectively.⁵

Developmental cysts are generally asymptomatic, but have the capacity to grow extremely large and cause cortical expansion and erosion.⁶ The most prevalent type of developmental cysts are dentigerous cysts followed by odontogenic keratocysts (OKC). Paget defined dentigerous cysts in 1863 as slow-growing odontogenic cysts of developmental origin that are most commonly correlated with the crown of an unerupted or developing tooth. The cysts comprising the crown of an unerupted tooth is also

connected to the tooth along the cervical region.⁷ The teeth most commonly involved are mandibular third molars, maxillary canines, and mandibular premolars.⁸ Dentigerous cysts often arise as a result of an accumulation of fluid between the remains of the enamel organ and the subjacent tooth crown shortly after the crown has fully formed. Dentigerous cysts expansion is associated with a subsequent rise in cyst fluid osmolality caused by the entrance of inflammatory cells and desquamated epithelial cells into the cyst lumen.⁹

The odontogenic keratocyst (OKC), originally reported by Philipsen (1956), is currently known as a keratocystic odontogenic tumor by the World Health Organization (KCOT).¹⁰ It is described as a benign uni or multicystic intraosseous tumor of odontogenic origin with a distinctive lining of parakeratinized stratified squamous epithelium and the tendency for aggressive, infiltrative activity.¹¹ The lesions grow mostly in the anteroposterior dimension, and they can grow extensively without severely deforming the jaw bone. The distinctive tendency for rapid development is caused by increased osteolytic activity of prostaglandin substances in the cell population of the cyst lining, and increased hyperkeratotic scale deposition in them cyst lumen, which results in a larger hydrostatic pressure differential.¹²

This case report features an unusual giant mandibular cyst that does not exhibit any of the standard characteristics of developmental odontogenic cysts.

CASE REPORT

A 54-year old female reported to Soedjati Soemodiardjo Purwodadi Hospital with a complaint of swelling over the left side of face for past 3 years showing facial deformity. On extraoral examination, a large painless swelling was seen which was hard

in consistency and non-tender about 2 x 7 cm in diameter showing facial asymmetry. The swelling has a diffuse border without ulceration and skin infiltration. It extends from anterior mandible to the left angulus. The anterior part of the mandible is palpable hard solid, the corpus to the left

mandibular angulus is palpable crepitation. Intraorally, the swelling was covered by normal mucosa extending from lower left second premolar to the angle of mandible with no ulceration (Figure 1).



Figure 1. Preoperative Clinical Condition



Figure 2. Preoperative Panoramic Radiograph

OPG revealed a cystic multilocular lesion extending from the mesial lower first molar of the mandible to the left angle of mandible. An impacted

paramolar supernumerary teeth was also present in the lower interdenal of lower left second premolar and first molar (Figure 2).

The proper treatment in most cases is careful cyst enucleation and removal of the impacted tooth. In view of the large size of the lesion, treatment plan of enucleation was made by taking the cyst capsule in toto and can be treated with decompression (marsupialization).^{13,14} Allowing for an accurate diagnosis and decompression. Then, the remaining capsule can be enucleated

without risking surrounding noble anatomical structures such as the inferior alveolar neurovascular bundle and adjacent vital teeth. Prognosis is excellent in most case, as seen in this case after 8 months healing, the patient felt that the mass in mandibular was increasing, there were not complaints of pain, and after orthopantomogram the lumen of cyst began to fill with new bone (Figure 3).



(a)



(b)



(c)

Figure 3. clinical condition 1 month after surgery (a), 5 months postoperative radiograph (b), 8 months postoperative radiograph(c)

DISCUSSION

Odontogenic cysts are the most common form of cystic lesions affecting the maxillofacial region. They are classified traditionally into a developmental group, including keratocysts and dentigerous cysts, and an inflammatory group, including radicular cysts. The odontogenic keratocyst (OKC) was first described by Philipsen (1956), is now designated by the World Health Organization as, a keratocystic odontogenic tumor (KCOT). It is defined as “a benign uni or multicystic, intraosseous tumors of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviors.” The percentage of OKCs versus other cysts of the jaws as given by different authors are as follows: Hjorting-Hansen et al. (1969) and Toller (1972) as 11%; Brannon (1976) and Payne (1972) as 9%; and Pindborg and Hansen (1963) as 7%. Growth is chiefly in the anteroposterior dimension, and the lesions may attain remarkable size without significantly deforming the jaw skeleton. The particular tendency to rapid growth is due to the higher

activity of the epithelial cells of the cyst lining, stimulating osteolytic activity of prostaglandin substances in the cell population of the cyst lining, and the higher accumulation of hyperkeratotic scales in the lumen of the cyst, resulting in greater difference in hydrostatic pressure.¹⁵

Odontogenic keratocysts are lesions that derive from the epithelial remains of the dental lamina, they have slow growth, they are aggressive as they are expansive and highly recurrent, they may or may not be associated with syndromes, they have a higher incidence in the 2nd decade of life and may have another peak. between 55-65 years; There is a certain predilection for the male gender, the differential diagnosis is broad both clinically and radiographically. It can be managed with conservative treatment (enucleation, curettage, cryotherapy, chelating agents) or aggressive management (bloc or marginal resection). The objective of the present study is to introduce silicone valve decompression as part of the treatment of odontogenic keratocysts, since there are not enough studies in this regard in Mexico.

We present a case of a patient with an Odontogenic Keratocyst, treated by the Maxillofacial Surgery service of the "La Raza" Medical Center, who underwent decompression with a silicone valve. The long-term result is presented by radiographic control after three years without reporting complications or recurrence.¹⁶

Dentigerous cyst is a common type of odontogenic cyst of the oral and maxillofacial region. It commonly occurs in the second or third decades and rare cases appear in the first decade of age. It is a developmental cyst associated with unerupted/impacted tooth. Radiographically, it appears as unilocular, radiolucent area along the cemento-enamel junction of the associated tooth. Larger dentigerous cysts may be treated with marsupialization, and smaller dentigerous cysts are treated with enucleation of the cyst and extraction of the associated tooth.¹⁷

Age and Sex Predilection 485 cases (57.2%) were observed in men and 362 cases (42.7%) were seen in women, with a male: female ratio of 1.34:1. The mean age was 28.2 years (range: 5-86 years), with 262 cases (30.9%) and 316 cases (37.3%) being diagnosed in the second and third decades of life, respectively. Radicular cysts were most frequently seen in the second and third decades, dentigerous cysts in the second decade, and odontogenic keratocyst in the third decade of life. Location Of the 847 cysts, 410 cysts (48.4%) were on the maxilla and 437 cysts (51.6%) were on the mandible. In maxilla, the anterior region was the most commonly involved region (271 cysts, 66.1%), whereas in the mandible the posterior region (219 cysts, 50.1%) and anterior region (218 cysts, 49.9%) nearly had equal prevalence. The inflammatory cysts (radicular or residual cysts) was

predominantly encountered in the anterior maxilla (59 cysts, 53.2%). Development cyst was most commonly seen in the posterior mandible (Dentigerous cysts: 50.8%, Odontogenic keratocyst: 55.56%).¹⁸

The etiology of odontogenic keratocysts may arise from proliferation of the dental lamina epithelium of the jaws, from basal cells of the oral epithelium, or from degenerated portions of enamel by the stellate reticulum, prior to the initiation of enamel apposition.¹⁹ Several studies have reported that the occurrence of odontogenic keratocysts (OKC) is associated with changes in tumor suppressor genes on chromosomes 2q22.3-q31.36. Tumor suppressor genes involved in OKC development are p16, p53, PTCH, and MCC. The pathogenesis of OKC involves a "2-hit mechanism" associated with loss of the allele at 2q22. The "2-hit mechanism" is a process of activating tumor suppressor genes. Inactivation of tumor suppressor genes such as p16, p53, and PCTH will cause inhibition of apoptosis that triggers the development of OKC. In addition to genetic factors, various studies have shown that cell cycle dysregulation and cell proliferation also play a role in the pathogenesis of OKC. Due to this condition OKC showed an increase in cell proliferation which resulted in aggressive growth of OKC. The proteins involved in the cell proliferation process are PCNA and Ki-67.²⁰

Jaw cysts are the most common cysts, because of the large amount of epithelial remnants left in the tissue after tooth formation. Jaw cysts are divided into two, namely non-odontogenic cysts and odontogenic cysts.²¹ The majority of cysts are small and do not cause swelling on the tissue surface. If there is no infection, then clinically the enlargement is minimal and well-

defined. Enlargement of the cyst can cause facial asymmetry, shifting of teeth and changes in occlusion, loss of associated or adjacent teeth, and displacement of the denture. Cysts that are located near the surface and have expanded into the soft tissue, often look bright blue in color and the mucous membrane covering is very thin. However, dystrophic calcifications can occur in long-developed cysts, causing the cyst to be not completely radiolucent in its internal structure. Cysts can be unilocular or multilocular.²¹

Keratocyst Odontogenic Tumor (KCOT) is a benign odontogenic tumor originating from epithelial tissue. Based on the histological picture, KCOT consists of three types, namely parakeratin, orthokeratin, and a combination of both. KCOT is a type of odontogenic tumor that is closely associated with unerupted jaws and teeth. Both types of tumors are very aggressive or have the potential to become neoplasms and tend to recur.²² The first clinical signs appear in the form of painless swelling, facial deformity, pain when the swelling affects other structures, spontaneous tooth loss, intraoral ulcers, and periodontal disease. Ameloblastoma is most commonly found on the posterior aspect and angle of the mandible, but can occur anywhere in the maxilla or mandible.²² Radiographically, KCOT appears as an irregular radiolucent, uni or multilocular, which generally has a thin, reactive sclerotic layer, and a smooth or scalloped margin and penetrates into adjacent bone. In this panoramic case, it appeared as a radiolucent, unilocular lesion without root resorption, a destructive cystic lesion in which all the cortical bone of the

TMJ and mandibular ramus had been resorbed.²²

Dentigerous cysts or follicular cysts are odontogenic cysts associated with crowns of impacted permanent teeth, unerupted permanent teeth, supernumerary teeth, odontomas, and rarely affect primary teeth. These cysts are often found in young adults, and adults, and are rarely found in children. The mandibular third molars and maxillary canines are common cases.²³ The radiographic appearance of a dentigerous cyst is a radiolucent symmetrical, unilocular, well-defined, cortical, and surrounds the crown of the impacted tooth. The slow, regular growth of the cyst gives dentigerous cysts a poorly demarcated sclerotic margin.²³ Local factors for the cause of dentigerous cysts can be associated with the development of crowns or permanent teeth but can also arise from remnants of the enamel epithelium. Cysts can grow to any size, and large cysts can be associated with painless expansion of the affected bone. Large lesions can cause facial asymmetry and can be potentially aggressive.²³

CONCLUSION

The management of an uncommon case of giant mandibular cyst using the enucleation method by removing the entire cyst capsule followed by decompression (marsupialization) and removing the associated tooth can result in a good prognosis in this case.

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