



Uncommon case report of maxillary sporadic odontogenic keratocyst

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ABSTRACT

The odontogenic keratocyst (OKCs) is a rare disease manifested in the maxillofacial region, mainly affecting the mandible rather than maxilla and is associated with the presence of a retained tooth. Most cases are asymptomatic, affecting males than women in their second decade of life and patients may present pain or soft tissue swelling in infected OKCs. Bony expansion and paresthesia of the lips are reported in fewer cases. The most common region in the maxillae is the third molar and cuspid region. The objective of this report is to present an uncommon clinical case of a 72 years old male with asymptomatic OKCs in the left maxilla region, presenting with bone lysis in the vestibular and palatine region with nostril alloy without compromising the teeth. Patient was treated with enucleation and peripheral osteotomy using a rotary instrument, today day without recurrence. A simple review of cases with maxillary keratocyst was made too.

Keywords: Odontogenic keratocyst, maxillary keratocyst, asymptomatic keratocyst, bone lysis, enucleation.

INTRODUCTION

According with the World Health Organization (WHO) odontogenic keratocysts (OKCs), earlier known as keratocystic odontogenic tumors, is defined

as «benign uni- or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified, has slow growth at the beginning, but with potential for aggressive, infiltrative behavior». ¹⁻⁴ There has been cases reports of OKCs in other nonosseous locations, which involve the gingiva, but mucosal, epidermal and even intramuscular sites. ³ OKCs presents as swelling with or without pain and occasionally paresthesia of the lower lip. ^{3,4} The cyst may reabsorb bone and cause a secondary teeth displacement with consequent malocclusion. The current literature cites a broad range of recurrence (0-62%), with most recurrences presenting within 5 years on average after a surgical treatment, if not removed completely. ^{3,5} It affects the mandible in a greater proportion than maxilla (60-75%). It is more common in the posterior ramus of the mandible, whereas in maxillae is in the third molar and cuspid region. ^{3,4,6-8} The frequency varies from 11 to 21% and it can appear at any age (8-82 years), but shows predominance in patients between 20 and 40 years with higher male prevalence (1.6:1). ^{3,4,6,8} It is one of the rare and distinctive developmental odontogenic cyst which from the dental lamina, it is named this way because keratin is produced by the cystic lining. ⁴ A Cyst is an abnormal cavity, lined with 6 to 10 cell layer parakeratinized stratified squamous epithelium, and a thin walled within the bone. It contains clear liquid or semi-liquid fluid with a cheesy material representing parakeratin produced by the luminal epithelium, mesenchymal and/or ectomesenchymal that are or were part of the odontogenesis process. ^{4,6} Under the microscope OKCs exhibits a wavy, corrugated surface with a prominent, palisaded and hyperchromatic basal

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cell layer, with epithelium that has a flat interface with the adjacent connective tissue.^{4,6} Treatment with marsupialization «decom-pression or cystotomy» also results in the loss of classic features, with the lining becoming reminiscent of the normal adjacent mucosa. The epithelium may strip away from the connective tissue in these cysts, therefore close examination of the biopsy container for material is recommended.^{1,4,8,9} Treatment options range from a conservative approach to extensive surgery that include enucleation with cryotherapy, marsupialization, decompression and segmental resection.^{4,6,9}

CLINICAL CASE

72 years old male was referred to the *Facultad de Odontología, Universidad Autónoma de Guadalajara* for routine evaluation. On physical examination there was an asymptomatic patient with preserved oral cavity, normochromic mucosa, and symmetric bone structure (*Figure 1*). Routine orthopantomography was requested and it demonstrated a radiolucent left maxilla region, molars and pre-molars (*Figure 2*). By computed tomography (CT) with 3D reconstructions she showed hypodense circular area (1.9 × 2.5 × 2.0 cm)

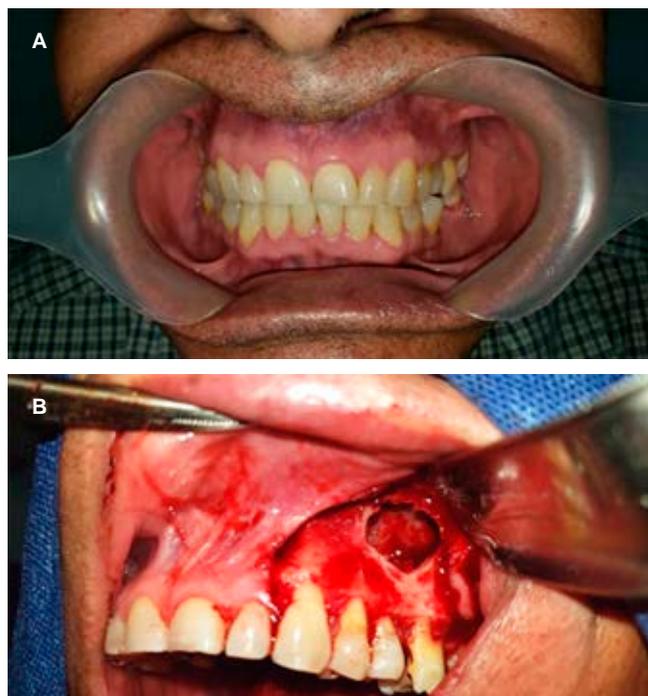


Figure 1: Intraoral of view. **A)** Mucosa coloration normal and bone structures are symmetric. **B)** Surgical resection of the Keratocyst, was confirmed bone lysis in the palatal region and communication with the ipsilateral nostril.

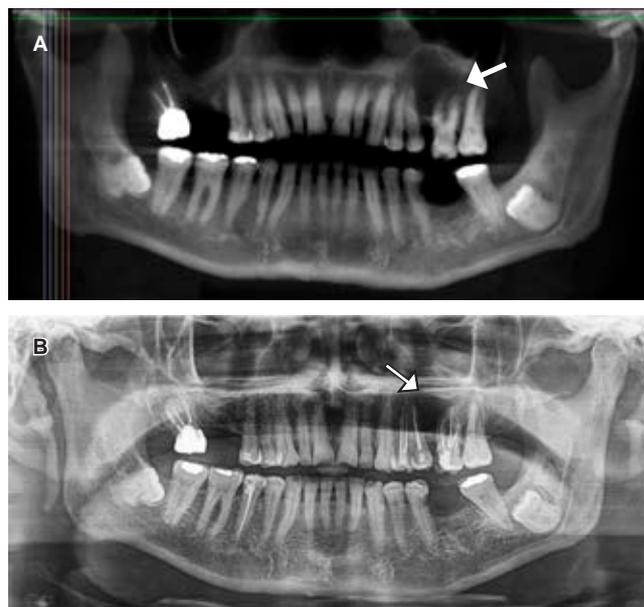


Figure 2: Orthopantomography. **A)** The arrow shows radiolucent area with a defined edge defined, **B)** Control orthopantomography at 6 months. The area occupied by the Keratocyst is in frank recovery.

with defined edges (*Figure 3A*) communicated with the nasal cavity, osteolysis in the palatine region and the anterior wall of the left maxilla (*Figure 3B*).

Treatment: the enucleation was done under local anesthesia with a combination of a peripheral osteotomy using the rotary instrument through a canine triangular mucoperiosteic flap in the second molar of the left maxillary. Osteolysis was found in the palatal region communicated with the ipsilateral nostril, previously observed on the 3D reconstruction (*Figure 1B*). Removal was performed and a sample was placed in 10% formaldehyde for laboratory histopathology examination. This technique was used since the complete removal of the cyst possessed no risks of complications from a dental and/or anatomical point of view. Furthermore, this method facilitated the comprehensive analysis of the lesion including its clinical, histopathological, and radiological aspects.

Presumptive diagnosis: due to location, clinical characteristics, radiography, absence relation between the retained tooth and patient's age, it is set a presumptive diagnosis of Giant Cell Central Granuloma or OKC.

Definitive diagnosis: the histopathology reports a cavity without apparent content, thin odontogenic epithelium lining without projections towards its capsule and detached in most sections of its fibro-

connective capsule. Flat stratified epithelium of 6 to 8 cell strata, hyperchromatic basal layer and para-keratinized surface were identified, with all these pathological features allow the diagnosis of OKC (*Figure 4*).^{1,6,7}

DISCUSSION

The database patient records of the Department of Maxillofacial Surgery of the Facultad de Odontología de la Universidad Autónoma de Guadalajara from June 2014 to November 2019 was reviewed. Recognition of 132 biopsies was found, two of them were odontogenic cysts (2.3%), one dentigerous and one radicular. However, as it shown in *Table 1*, four articles were located in the review of works analyzing odontogenic cyst cases in Mexican population (three carried out in Mexico City and one in San Luis Potosi City). Of the total Biopsies 9,673, of which 1,266

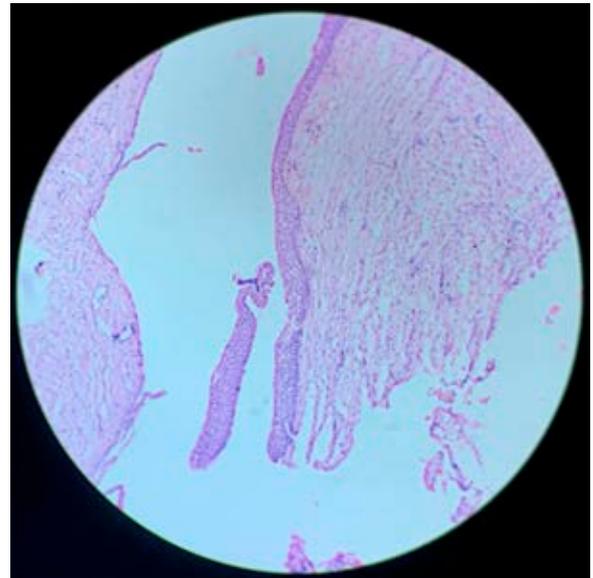


Figure 4: Ontogenic para-keratinized lining epithelium with hyperchromatic basal stratum. Hematoxylin-eosin staining (10x).

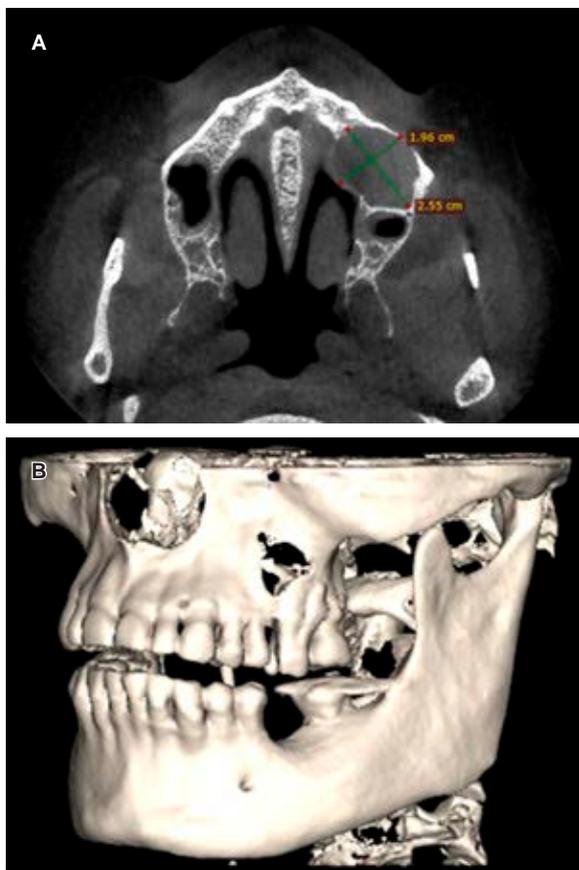


Figure 3: Computed tomography scan cut axial facial (CT) shows. **A)** Radiolucent circular area in left maxillary sinus of 1.96 × 2.55 cm. **B)** Bone destruction in the anterior wall of the left maxillary sinus.

(13.1%) were diagnosed as odontogenic cysts and 306 cases (24.2%) were OKCs.¹⁰⁻¹³ In Mexican data base (*Table 1*), both percentage of odontogenic cysts (*Table 2*) and the slight preponderance of OKCs in men were similar to those described in the literature.^{3,4,6,8} On the other hand, the percentage of OKC was different in the Mexican population (24.2%) than the other population, in which the percentage varies from 1.69 to 18.6%. It is mostly probable that the difference in the percentage of OKC between the review of the other population, Mexican and our database, could be due to the difference in years analyzed or the number of cases treated per year. Also, it could even be due to diagnostic methods, even if it is not described, it could have involved some genetic component, since the variability between one study and the another is quite wide (*Table 1*). Therefore, this represents a rare case, if the whole clinical picture is considered as a whole, since OKC according to the literature occurs at all ages with peak incidence in the 2nd and 4th decade of life,⁴ even in Mexican population.²¹ Nevertheless in our clinical case, the patient was 75 years old and the oldest person of the all cases we knowledge with OKC in the maxilla, further OKC in maxillary region are rare with a low frequency in the left region (*Table 2*),^{4,6,7} as Cadena-Anguiano described it in Mexican population study.¹⁴ However, the maxillary involvement owns a greater and increased threat, due to proximity to

vital structures such as maxillary sinus, orbital floor, and infratemporal fossa.¹⁵ Medline, Google Scholar and Google search was performed using the MeSH term «keratocyst», combined with the word «maxilla» and «case report». We detected 32 cases of maxilla OKCs, includes our case (Table 2), and it is noted that 34.4% of all cases described were from India, 15.6% from Brazil and 9.4% (three cases, including ours) from Mexico. In these studies, 46.9% were in the left maxilla, 65.6% without pain, 43.7% without retention of dental organ and 53.1 % without bone lysis. Of the 32 cases, only 3 cases (9.4%) (Including ours) did not show any symptoms, however, they developed bone lysis (Table 2).

On the other hand, odontogenic cysts constitute a group of maxillary lesions, which cause their destruction and due to its aggressive potential, as well as its high recurrence rate, surgical treatment has been controversial challenge. The decision will depend on multiple factors, mainly anatomical location, size and compromised anatomical structures with the objective of eradicating the lesion and decrease complications as small as possible.¹⁶⁻¹⁸ Considering our patient OKC, it caused bone lysis in the mouth palatine region as well as communication with the nostril, conservative treatment decision was

made, in order to avoid damage to the nostril, as well as the palatine region. Based on the fact that the OKC is a lesion that has a high recurrence rate^{3,4,6,8} and to reduce this risk, it was decided to perform peripheral osteotomy with a rotary instrument after enucleation in order to decrease the recurrence rate. After resection of the lesion, taking into account the OKC as a presumptive diagnosis, it is decided to carry out adjuvant therapy discarding the Carnoy solution and Cryotherapy to avoid damage to the soft tissues of the palatine region and nasal fossa, in addition a radiographic surveillance is established every 6 months, the first one (Figure 4) shows adequate bone healing and decreased defect, and no evidence of recurrence. The cause of recurrence is not entirely clear, but it is attributed to the thin and friable layer of connective tissue of the cyst which results in incomplete removal or the presence of satellite cysts.^{9,53}

CONCLUSION

Although the OKC is an entity with a high recurrence, it should be considered that aggressive treatment is not advisable in all cases, due to factors such as commitment to anatomical structures and

Table 1: Odontogenic cysts in Mexican and other countries population.

Year period	Per year	Biopsies		Odontogenic cysts type				City, ref.
		Total	O. cysts n (%)	OKCs n (%)	Dentigerous n (%)	Radiquar n (%)	Other n (%)	
5	26	132	3 (2.3)	1*	1	1	0	Gdl. City, Mex. 2020*
18	70	1,266	103 (8.1)	34 (33) M (50)	58 (56)	5 (4.9)	6 (5.9)	SLP City, Mex. ¹⁰
6	116	700	75 (10.7)	30 (40) M 13 (43.3)	21(28)	ND	24 (32)	Mex. City, Mex. ¹¹
21	210	4,410	856 (19.4)	184 (21.5) M 108 (58.7)	283 (33)	342 (39.9)	47 (5.6)	Mex. City, Mex. ¹²
10	386	3,865	304 (7.9)	57 (18.7) M 34 (59.6)	108 (35.5)	ND	139 (45.7)	Mex. City, Mex. ¹³
		9,673	1,266 (13.1)	306 (24.2) M 155 (50.5)	45.7 (37.2)	348 (27.5)	216 (17)	Mexican ¹⁰⁻¹³
11		2,370	-- (10.7)	---	122 (5.2)	---	93 (3.9)	American ¹⁹
04	---	475	340 (71.6)	23 (6.8)	77 (22.6)	216 (63.5)	24 (7.1)	Turkey ²⁰
10		8,563	1,518 (17.7)	282 (18.6)	366 (24)	506 (33.2)	126 (8.4)	Iranian ²¹
58		1,305	-- (18.5)	22 (1.69)	123 (9.4)	68 (5.2)	---	Australian ²²
10		---	125 (---)	17 (13.6)	28 (22.4)	76 (60.8)	4 (3.2)	Indian ²³
12		---	406 (11.26)	61 (15.02)	57 (14.04)	186 (46.06)	102 (24.9)	Brazilian ²⁴

M = male, O. cysts = odontogenic cysts, OKCs = keratocysts, SLP = San Luis Potosi.

* This case.

Table 2: Odontogenic keratocysts no syndrome in the maxillary region.

	Age (yr)/gender	Side	Symptom			Tumor	Tooth	Bone lysis	Country, ref.
			Time	Swelling	Pain				
1	6/F	Left	--	+	-		-	-	Japan ²⁴
2	7/M	Right	60 d	+	Continuous non radiating		+	-	India ²⁵
3	8/M	Right	1 yr	+	-		+	+	India ²⁶
4	9/F	Both	---	+	-	Solid	+	-	USA ²⁷
5	14/F	Left	---	-	-		-	+	Brazil ²⁸
6	14/M	Right	14, yr	+	-	Purulent fistula	+	-	Brazil ²⁹
7	15/M	Right	60 d	-	Tooth painon		-	+	India ³⁰
8	15/F	Left	90 d	+	Moderate, radiating		+	+	India ³¹
9	15/M	Right	3 yr	+	+	Pus discharge	+	+	India ³²
10	17/M	Left	7 d	-	Persistent headache	Milky White Liquid	+	-	Brazil ³⁰
11	18/F	Right	4 yr	+	Light recurring	Cheesy fluid	-	-	Mexico ³⁴
12	18/F	Left	-	-	-		-	-	Korea ³⁵
13	19/M	Right	-	-	-		+	-	Mexico ³⁶
14	21/M	Both	7 d	+	Continuous, non-radiating		-	+	India ³⁷
15	22/F	Left	6 m	+	Intermittent	-	-	-	UK ³⁸
16	22/F	Left	-	-	-		-	+	India ³⁹
17	23/M	Left	90 d	+	-		+	-	Spain ⁴⁰
18	26/F	Right	1 yr	+	-		+	-	Iran ⁴¹
19	27/F	Right	-	-	+		+	+	Brazil ⁴²
20	28/F	Right	30 d	+	-		+	-	Brazil ⁴³
21	32/M	Left	90 d	+	-		+	+	Spain ⁴⁴
22	32/M	Left	60 d	+	-		+	+	India ⁴⁵
23	34/M	Left	--	+	+	Green, cheesy Fluid discharge	-	-	USA ⁴⁶
24	36/F	Right	30 d	-	-	Tumefaction, straw colored Fluid	+	+	Kosovo ⁴⁷
25	39/F	Right	0.5 yr	+	+	No fluid	+	-	Japan ⁴⁸
26	45/F	Right	0.5 yr	+	-		-	+	India ¹⁵
27	46/F	Left	30 d	+	-		-	-	India ⁴⁹
28	54/M	Left	ND	-	--		-	+	Korea ⁵⁰
29	57/M	Left	ND	-	-		+	+	Korea ⁵⁰
30	64/F	Right	30 d	-	Intermittent non radiating		+	-	India ⁵¹
31	72/M	Right	Several months	+	-	-	-	-	Israel ⁵²
32	75/M	Left	-	-	-	Solid	-	+	Our case, Mexico
	n = 32	Left	Min.-	(-) n = 12	(-) n = 21		(-) n = 14	(-) n = 17	
	M	n = 15	max.	37.5%	65.6%		43.7%	53.1%	
	n = 16	46.9%	7 d-4 yr						
	50%								

F = female, M = male, MS = maxillary sinus, yr = year, m = months, d = days, RE = recurrence after surgery, ND = no data.

possible complications should be considered, however radiographic controls are advised periodically.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the

patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: None.

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