The odontogenic keratocyst of the maxilla

A review of the literature and analysis of cases treated in private practice in a thirty-five year period

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Abstract

German: Die Gesamtzahl odontogener Keratozysten wurde in einer Privatpraxis über einen Zeitraum von 1986 bis 2021 ermittelt. Die Gesamtzahl betrug 34. Eine Unterteilung erfolgte in Unterkiefer- und Oberkieferkeratozysten. Insgesamt wurden zehn in diesem Zeitraum behandelte Oberkieferkeratozysten (29,41 %) weiter analysiert. Es erfolgte eine umfangreiche Durchsicht der aktuellen Literatur.

Introduction and review of the literature

The odontogenic keratocyst is a cystic lesion which in the majority of cases is found in the mandibular posterior region. Involvement of the maxillary sinus by an odontogenic keratocyst has been reported as a rare finding.[1] Shear stated that 75% of cases occurred in the mandible and only 25% occurred in the maxilla.[2] This agreed with most of the reports quoted by other authors, which ranged from 77 to 69% in the mandible. Neville, Damm, Allen and Bouqout[3] showed the relative distribution of keratocysts. About 30% of lesions occured in the maxilla whilst the vast majority were in the ramus and angle areas of the mandible. These figure were approximately the same as found by Shafer et al.[4]
Nayak et al.[5] reported that various names have been given to this entity. These included a cholesteotoma in 1926, primordial cyst first described by Robinson in 1945, the odontogenic keratocyst as described by Philipsen and Pindborg in 1956 and by Pogrel in 2012 as the keratocystic odontogenic tumour.[6] The name changed back to odontogenic keratocyst when the World Health Organization (WHO) Classification of Head and Neck Tumours was published in 2017 and was highlighted by Borgese et al.[7] where the different terminologies were considered. From 2017 to the present, the term odontogenic keratocyst was again preferred but the term keratocystic odontogenic tumour was also acceptable.[7, 8]
All publications reviewed emphasized that the odontogenic keratocyst, as it is termed today, was a locally benign very aggressive lesion. The lesion could recur and various methods of removal of the tumour were advocated in order to minimise recurrent lesions.[1–5, 7, 9, 10]
June-Ho Byun et al.[10] considered the immunochemical analysis of the specimens from two patients who had more aggressive maxillary odontogenic keratocysts. The chemical analysis showed p53 & p63 strong expression, but B-cell lymphoma 2 (BCL2) and MK167 (KI67) showed moderate or weak expression. They also showed the associated protein BCL2 and BAX was almost negative. They deduced that there may be increased antiapoptotic activity which could result in the enlargement, high recurrent rate, and aggressive behavior of the lesion.
Malignant transformation of this lesion and transformation either into squamous cell carcinoma or invasive ameloblastoma was reported by Jalani et al.[9] and Ahmadian et al.[11] The rarity of odontogenic keratocyst also as keratocystic odontogenic tumour of the maxilla was highlighted by Al-Bodbaij et al.[12] and Silva et al.[13] and Sampieri et al.[14]

Method

Ten patients with histologically diagnosed maxillary keratocysts of a total

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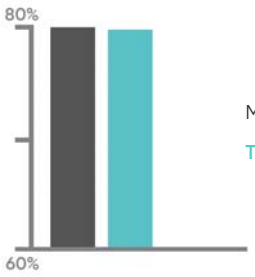
Porositätsanalyse[1]



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[1]Ref Human trabecular bone: Renders GA, Mulder L, van Ruijven LJ, van Eijden TM. Porosity of human mandibular condylar bone. J Anat. 2007;210(3):239-248.
Ref THE Graft: Lee JH, Yi GS, Lee JW, Kim DJ. Physicochemical characterization of porcine bone-derived grafting material and comparison with bovine xenografts for dental applications. J Periodontal Implant Sci. 2017;47(6):388-401.

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Number	Male/ Female	Age	Location	X-Ray	Wisdom or no wisdom	Left/ Right	Size	Date reported	Reported as	Treatment	Follow-up	Recurrence	Posterior wall of maxilla disruption on panorex
1	Female	26	Sinus posterior	Panorex PA tooth PA maxilla 15° occipito mental lateral skull	No wisdom	Left	20 x 10 mm	07/02/ 1991	Primordial cyst	Enucleation only	No	No	Yes
2	Male	51	Sinus posterior	Panorex CT scan	No wisdom	Left	20 x 10 mm	30/12/ 2010	Odontogenic keratocyst	Enucleation & BIPP	Yes 3 years	No	Yes
3	Male	39	Whole sinus	Panorex CT scan	Wisdom malformed	Left	20 x 10 mm	21/09/ 2012	Odontogenic keratocyst	Enucleation only	Yes 3 years	No	Yes
4	Female	51	Sinus whole	Panorex CT scan PA tooth	No wisdom	Right	Not stated	25/09/ 1992	Primordial cyst	Enucleation & BIPP	No	Unknown	Yes
5	Female	21	Whole sinus	Panorex CT scan PA tooth	Wisdom 18 displaced	Right	Not stated	31/05/ 2000	Odontogenic keratocyst	Enucleation & BIPP	No	Unknown	Yes
6	Male	31	From 14 to posterior maxilla	Panorex CT scan	No wisdom	Right	25 x 11 mm	15/05/ 2000	Odontogenic keratocyst	Enucleation & BIPP	Yes 10 years	No	Yes
7	Female	53	Whole sinus	Panorex CT scan PA tooth 27 CBCT	No wisdom	Left	25 x 13 mm	16/03/ 2020	Keratocystic odontogenic tumour	Enucleation & BIPP	Yes 2 years	No	Yes
8	Female	38	Anterior sinus	Panorex PA Occlusal plane maxilla	No wisdom	Anterior Sinus	10 x 5 mm	05/06/ 1998	Odontogenic keratocyst	Enucleation only	No	Unknown	No
9	Male	44	Left maxillary antrum whole	Panorex CT scan	No wisdom	Left	10 x 10 mm	05/10/ 1992	Odontogenic keratocyst	Enucleation & BIPP	Yes 4 years	No	Yes
10	Female	26	Left antrum posterior	Panorex	Wisdom 28 displaced	Left	20 x 10 x 20 mm	10/09/ 1996	Odontogenic keratocyst	Excision only & wisdom	Yes 1 year	No	Obscured by wisdom

Fig. 1: Parameters used in the analysis of 10 maxillary keratocysts.

of thirty-four (34) were treated in the period 1987 to 2021 in private practice, and not from archival records and institutions. This appeared to be just about the largest series of maxillary keratocysts in private practice recently reported. The lesions were all operated by one surgeon and diagnosed by oral pathologists.

The maxillary keratocysts were analysed according to 13 different parameters (Fig. 1). These included gender, age, location, the radiology available, the presence or absence of wisdom teeth, the size and site as well as the histological date reported as well as the name given to the lesion. Furthermore analysis of the treatment, follow up and recurrence was noted.

The presence of disruption of the posterior maxillary sinus wall on an orthopantomograph of the posterior maxilla and was of special interest to surgeons and dentists who encountered this finding in their practices.

Results

Gender

In the 10 maxillary keratocysts found, 6 were found in females and 4 in males (Fig. 2).

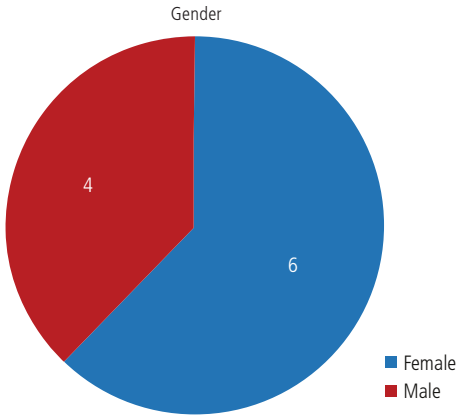

Fig. 2: Gender distribution of 10 maxillary keratocysts.

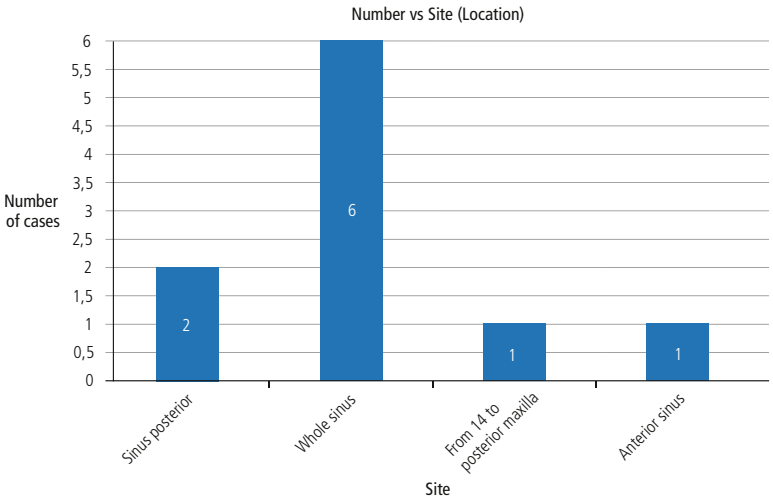

Fig. 3: Site distribution of 10 maxillary keratocysts.

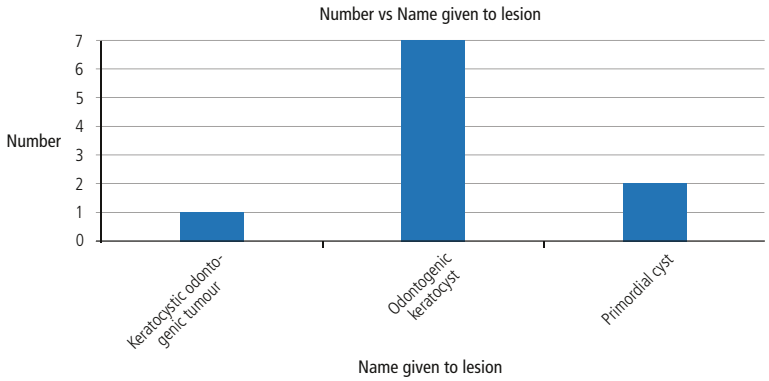

Fig. 4: Terminology used in histology reports.

Site and side

Of the 10 patients, 9 had keratocysts which filled most of the maxillary sinus. Only 1 was in the anterior area of the maxilla, 6 keratocysts occurred in the left sinus and 3 on the right hand side (Fig. 3).

Size and terminology

The size of the lesions as reported in the histological reports showed that they varied in size from 65 x 19 mm to 10 x 5 mm with the average size approximately 25 x 12 mm. Of the 10 maxillary keratocysts reported, only 2 were reported as primordial cysts. This occurred in 1991 and 1992 when these entities were known as primordial cysts. Only one lesion was reported as a keratocystic odontogenic tumour in 2020 when the terminology had changed. According to the WHO classification, 7 lesions were reported as odontogenic keratocysts (Fig. 4).

Treatment

All 10 lesions were treated by enucleation. Five were treated with enucleation only and five treated by enucleation with placement of BIPP (Bismuth Iodoform Paraffin Paste) gauze sutured in place and the cavity closed and which was later removed. Thorough ostectomy of the surrounding bone was performed in all cases. None of the lesions were treated by marsupialisation or enucleation following the use of Carnoy's Solution. All the specimens were sent to oral pathologists for histological examination.

Radiographs submitted

A variety of radiographs were sent by the referring practitioners prior to treatment (Fig. 5). All included orthopantomograph (Fig. 6) as well as others. A periapical of the tooth (Fig. 7), PA of the maxilla, occipito mental, lateral skull radiographs and CT scans (Fig. 8–10) as well as a cone beam CT were available. It was noted that in all the cases an orthopantomograph (Panorex) was available. In 9 of the 10 cases excluding the odontogenic keratocyst occurring in the anterior maxilla the posterior wall of the maxillary sinus had been eroded and completely disappeared.

Histological features

The histological features of the 10 maxillary odontogenic keratocysts were typical of all keratocysts found in the maxilla or mandible. The histological features of the odontogenic keratocyst typically consisted of a thin wall structure often difficult to enucleate. The lining was typically of stratified squamous epithelium about 6 to 8

layers thick with palisaded basal cell layer. Separation of the epithelial from the underlying connective tissue was often seen and was one theory for the difficulty of the removal and subsequent recurrences. The cyst contents could range from cheesy to thin in consistency (Fig. 11).

Treatment

All operations were performed under general anaesthesia with nasoendotracheal intubation.

Access to all of our maxillary keratocysts was via a Caldwell-Luc approach which gave excellent exposure to the lesion. The 10 maxillary keratocysts were treated with either enucleation only or enucleation with placement of BIPP (Bismuth Iodoform Paraffin Paste) gauze which was sutured in the cavity and subsequently removed. Peripheral ostectomies were performed in all cases to minimise recurrences. The use of Carnoy's Solution or decompression was not employed because of possible damage to adjacent structures.

All specimens were sent to oral pathologists for histological examination.

Recurrences and follow-up

Recurrence has been reported as a feature of all odontogenic keratocysts and not only of maxillary keratocysts. Accessibility and the large nature of maxillary keratocysts could result to their recurrence even years after removal.

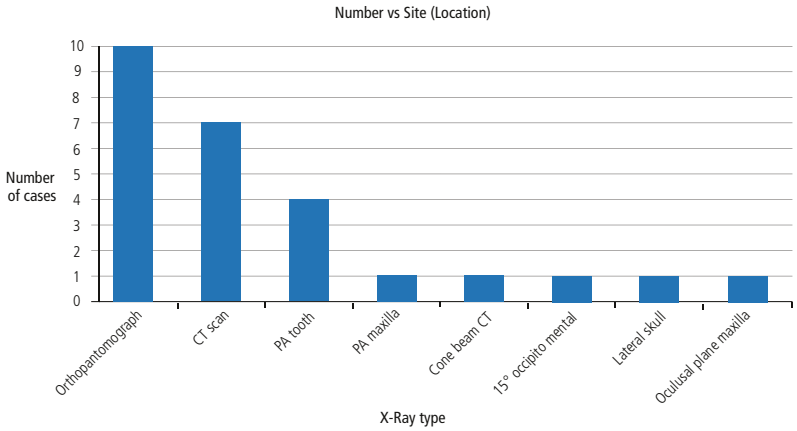

Fig. 5: Radiographs of various types sent by referring doctors.

Follow-up of our series of the 10 maxillary keratocysts ranged from 0 to 10 years. Four of the 10 patients failed to return after the surgery with follow-up of the remaining 6 patients varying from 1 to 4 years.

Recurrence was not found in any of our patients following either treatment described but always using peripheral ostectomy.

Discussion

Maxillary keratocysts were a rare entity as described in all of the literature reviewed. Our 10 cases of maxillary keratocysts showed a female predominance of 6 of the 10 patients which was an unusual finding.

A variety of radiographs were submitted with patients referred for pain and/or swelling of the sinus or even suffering toothache or tooth mobility. Periapical radiographs or orthopantomographs could easily be taken in the dental office and were often the first indication of a problem in the maxillary sinus. In all but one case where the keratocyst was in the anterior maxilla, the posterior wall of the maxillary sinus was eroded on radiograph.

No wisdom teeth (3rd molar teeth) were found in 7 of our 10 cases. Only 3 cases showed displaced or malformed wisdom teeth and therefore the presence of wisdom teeth within the lesion was variable.

In all our cases, except for the maxillary odontogenic keratocyst occurring in the incisor area most of the maxillary sinus was filled with the lesion. The extensive size of the lesion measuring 20 mm³ on average resulted in difficulty of removal of the lesion.

Peripheral ostectomy was performed in all patients and the placement of BIPP gauze in some patients was used in order to minimise recurrences.

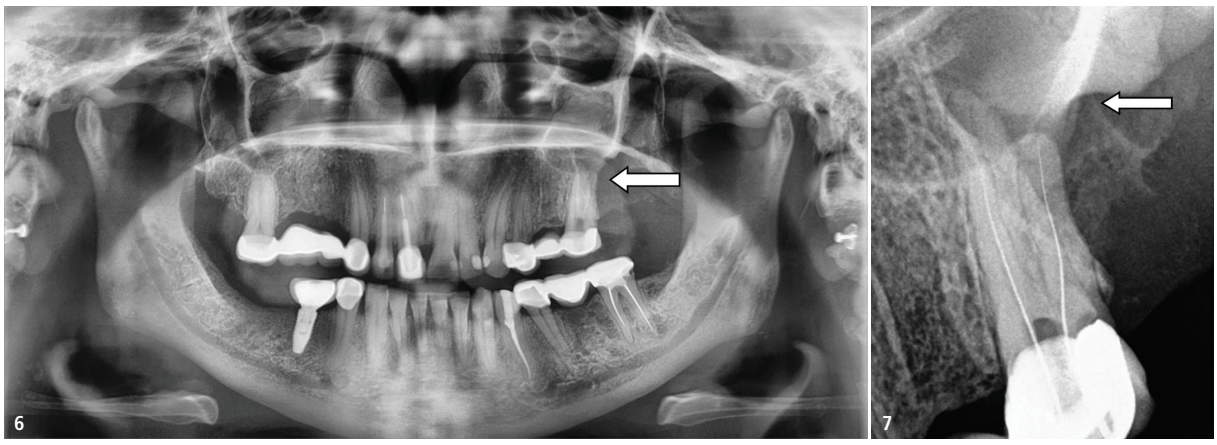

Fig. 6: Orthopantomograph showing loss of posterior antral wall on the left side in a patient with a large odontogenic keratocyst. – **Fig. 7:** Periapical showing loss of posterior antral wall in maxillary odontogenic keratocyst.

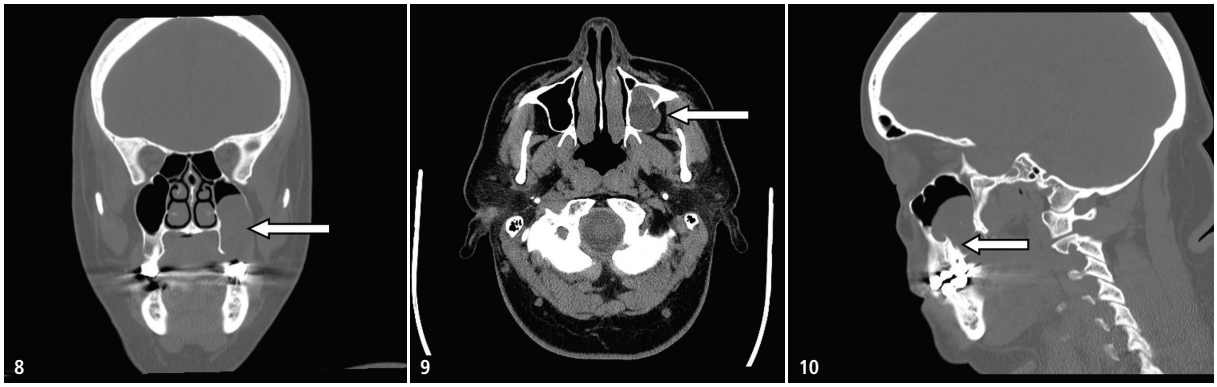

Fig. 8: Coronal CT of large maxillary keratocyst destroying left maxillary antrum. – **Fig. 9:** Axial CT of large maxillary odontogenic keratocyst occupying left sinus with destruction of antral wall. – **Fig. 10:** Sagittal CT view of odontogenic keratocyst in the antrum with bone destruction.

No recurrence of the maxillary keratocysts treated in this way was found in our study.

Conclusions

The maxillary odontogenic keratocyst was relatively rare and accounted for about 30 % of the keratocysts seen in our practice from the period 1986 to 2021. All were very extensive and occupied most of either the left or right maxillary sinuses and none were seen bilaterally. Careful dissection and removal followed by peripheral ostectomy and the placement of BIPP gauze with subsequent removal in some cases resulted in no recurrences of lesions which is notable of odontogenic keratocysts in general.

All the lesions were sent to oral pathologists for histological examination.

It is prudent for the dentist or surgeon to check on an orthopantomograph whether the posterior wall of the maxillary sinus was eroded before further radiological investigations in the form of either a CT scan, cone beam CT or MR were undertaken when the full extent of the lesion could be ascertained.

It is recommended to follow up these patients on an almost annual basis to check for recurrences.

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Conflict of interest

There is no conflict of interest.

References

All references (1–14) can be accessed in the electronic form of this paper and via the following link: qr.oemus.com/9751

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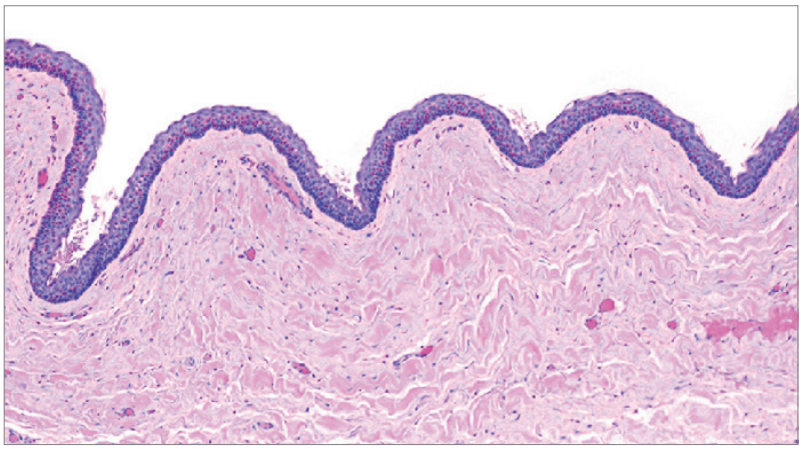

Fig. 11: The thin epithelial lining of the cyst showing palisading of basal cells and separation from the surrounding fibrous tissue that contained an inflammatory cell infiltrate typical of an odontogenic keratocyst (100x).

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Literatur

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