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RESEARCH ARTICLE

MANAGEMENT OF MISDIAGNOSED KERATOCYSTIC ODONTOGENIC TUMOR – A CASE REPORT OF 2 CASES

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ABSTRACT

Odontogenic keratocyst of the mandible is a challenging pathology to treat in Oral and Maxillofacial surgery. Secondary infections in large Odontogenic Keratocyst can lead to histopathological misdiagnosis. In such cases final diagnosis is usually made after complete surgical enucleation. We faced similar situation in two cases which are reported in the article.Enucleation with open packing was used for their treatment and has been found to be a good conservative approach in its management.

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INTRODUCTION

Management of OKC (odontogenickeratocyst) has been a controversial issue since the time PHILLIPSEN coined the term in 1956. Various treatment modalities have been used in literature for its management but still there is little or no literature on how to manage an initially misdiagnosed OKC. Although OKC has such distinctive histopathological features that microscopic diagnosis is rarely problematic, there are instances in which it is difficult to confirm the true nature of this lesion. This happens particularly when the cyst has become inflamed or infected. This article is about an alternative method for management of suchinitiallymisdiagnosed OKCs.

Report of cases

Case I

A 20 year old male complained of pain in mandibular anterior region for last 5 days with pus discharge since 3 days.

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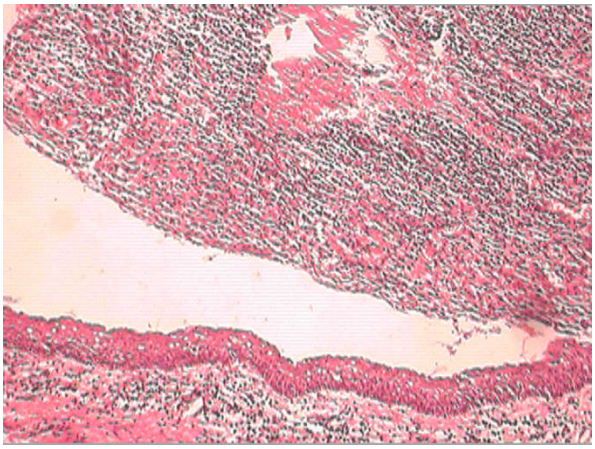
Patient gave a history of trauma to the same region 5 years back and extraction of right lateral incisor 4 years back. OPG X- ray revealed a large non corticated radiolucency extending from 2nd premolar on right side to the first premolar on left side of mandible suggesting a cystic lesion. The border was irregular, the right canine was displaced laterally and right lateral incisor was missing. An incisonal biopsy was performed (Figure 1a and 1b). The histopathological report suggested an infected radicular cyst. Complete enucleation was done under local anesthesia and primary closure done. The complete enucleated lining was sent for histopathological examination. The excisional biopsy report however showed odontogenickeratocyst (Figure 1c) (parakeratinized type). At this time a decision was made for open packing (iodoform gauge) of the lesion by creating a labial window. Patient visited regularly for change of iodoform dressing and OPG X-rays were taken at regular intervals of 3 months. A dramatic reduction in the size of the lesion and remodeling of the adjacent bone was noted in an OPG at 9 months after the initial surgery. At the 12 months follow up the OPG showed complete bone healing (Figure 2 a, 2b and 2c)

Case II

A 16 year old girl presented with pain and swelling in left mandibular region for last 15 days and pus discharge for the last 3 days.

Case 1. Inusional biopsy performed

Case 1. Orthopantomogram



Case 1. Parakeratinized epitheliumin excusional biopsy

Case 1. Complete healing at 12 months

OPG revealed a large radiolucent lesion extending from right second premolar to left second molar. Incisional biopsy was performed and reported as an infected odontogenic cyst (Figure 3a,3b).

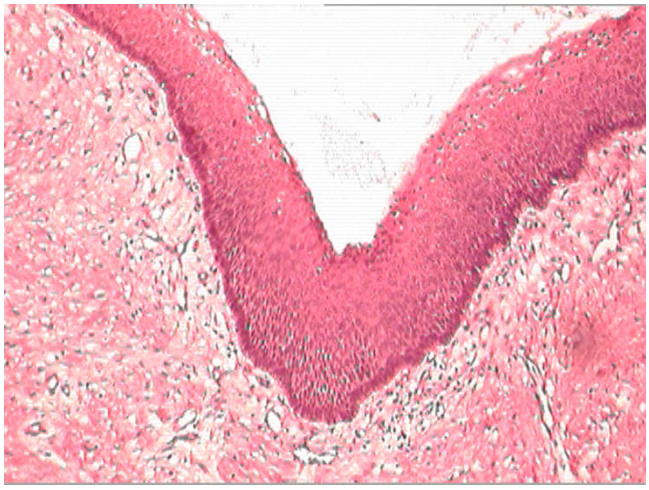
Cae 1. Complete bone healing at 9 months

Case 1. Complete bone healing at 12 months

Case II. Incisional biopsy done

Case II. Orthopantomogram shaory cystic lesion

The lesion was completely enucleated along with primary closure under LA and was sent for biopsy. The excisional biopsy report showed the lesion to be Keratocystic odontogenic tumor (Figure 3c) (parakeratinized type). At that time two windows were made on buccal and labial mucosa and cystic cavities packed with iodoform gauge which were regularly changed. The open packing continued for 2 months after which the bony cavities were completely filled up and only very small self cleansable pouches were left.



Case II. Excisional biopsy reveals parakeratinized epithelium

Case II. Complete healing at 1 year

Case II. Complete bone healing at 1 year

OPG X rays were taken every 3 months. In one year the lesion showed complete bone healing (Figure 4a and 4b).

DISCUSSION

Management of OKC is a highly debated lesion since the time PHILLIPSEN coined the term in 1956. Various treatment modalities have been used in literature for its management but still there is no consensus. There is very less or no literature on how to manage an initially misdiagnosed OKC. Although OKC has such distinctive histopathlogical features that microscopic diagnosis is rarely problematic; there are instances in which it is difficult to confirm the true nature of this lesion. This happens particularly when the cyst has become inflamed or infected. In presence of infection and inflammation, the cystic epithelium undergoes metaplasia to stratified squamous nonkeratinized epithelium (Maeda *et al*., 1990).

If only incisional biopsy is taken from such a lesion true diagnosis will be revealed only if tissue specimen is obtained from non inflamed or uninfected area. If the biopsy is not from a representative area, these cysts are misdiagnosed as any other infected odontogenic cysts. Many a times these lesions are enucleated like any other odontogenic cyst and it is only after the entire specimen is searched for evidence of typical OKC lining in areas without inflammation that a true diagnosis can be made.

The inflammed cystic epithelium which is difficult to diagnose is paradoxically the one having a higher proliferative fraction than the non inflamed OKC lining (Da Paula *et al*., 2000). Therefore, when OKC is suspected, it is important to search the entire specimen for evidence of typical OKC lining in areas in which there is no inflammation. Various surgical modalities have been mentioned in different literatures for the treatment of OKC, each having advantages and disadvantages of their own. The conventional options include (Phillipson, 1956) enucleation with curettage, (Maeda *et al*., 1990) enucleation and peripheral ostectomy and (Williams, 1194) enucleation with chemical cauterization (4) osseous resection without (rim ostectomy/ marginal resection) or with (segmental resection) continuity defects (Williams, 1194; Betanieh, 1998) (Ogden, 1992) marsupialization. But once the enucleation is done treatment options 1, 2, 3 and 5 do not practically exist and we are left with only one option that is osseous resection. As in both cases the lesion involved a significant part of mandible, resection would have resulted in severe esthetic and functional deformity and requirement of extensive reconstructive procedure. Although associated with 0% recurrence, resection of large part of jaw especially in young patients results in lots of psychological trauma and affects their job and marriage prospects.

We in our institute faced similar problem in two patients with large cystic lesions in mandible. Incisional biopsy in both cases was infected odontogenic cyst. Both these cysts were completely enucleated based on incisional biopsy and only when the whole cystic lining was carefully evaluated histopathologically the final diagnosis of Keratocystic Odontogenic Tumor was made. Both the patients were young and did not give consent for resection. At this time decision was made to create surgical windows on the buccal aspects of lesions to start open packing, expecting to achieve the advantages provided by decompression and marsupialization.
There are few reports in the literature of decompression or marsupialization of the OKC being performed for complete cure. These procedures are associated with following advantages –

- Simple method
- Less risk to adjacent vital structures like inferior alveolar nerve, teeth, maxillary sinus and nasal cavity (Eyre, 1985; Brondum *et al*., 1991 and Marker *et al*., 1996).
- After marsupilization the metaplasia of cystic lining to normal oral mucosa takes place when exposed to oral environment making it less aggressive or
- The cystic lining undergo into a creeping substitution by normal oral mucosa from the edges of the lesion
- Protein Bcl-2 which is an apoptotic protein did give consistently positive results in our hands in the keratocysts lining but was negative in normal oral mucosa and in the lining after decompression (Ogden, 1995)

The procedure that was carried out in 2 of our cases was open packing of the cyst through the windows made in labial and buccal wall. We used iodoform gauze for packing. It was prepared by soaking roll gauze in a mixture of glycerin and iodoformpowder. Before packing, the cavities were irrigated initially by saline and hydrogen peroxide and in the end it was irrigated by injection metrogyl. Packing was changed every 3rd day. Patients were kept on Chlorhexidine mouthwash twice a day throughout this period. Once the cavity size reduced considerably, we took incisonal biopsy from the base of residual lesion which now represented the base of cystic cavity. Histopathological evaluation of this tissue confirmed absence of any residual pathology.

RESULTS

Both were single cysts and none was syndromic although both were multilocular. In both cases the keratocyst resolved completely, both clinically and radio graphically.

CASE 1:

Initial size = 7.5 x 2.5 cm (as recorded from OPG)

Time taken for complete resolution:

- Clinically: 3 months
- Radiographically: 12months

Follow up period: 18 months

CASE 2:

Initial size = 6.5 x 2.5 cm (as recorded from OPG)

Time taken for complete resolution:

- Clinically: 7 months
- Radiographically: 12 months

Follow up period: 16 months

Teeth adjacent to the cyst or within the cystic cavity were extracted and root canal treatment with respect to 37 and 36 in 2nd case.

Both cases were of parakeratinized variety histo-pathologically. The cystic lining was enucleated completely. In both the cases the histological material obtained from base of defect after complete resolution, revealed no sign of cystic remnants, daughter cysts or budding of the basal layer of the epithelium.

The procedure of enucleation and open packing is associated with following advantages and disadvantages

Advantages

- With enucleation and open packing we are still getting the advantage of decompression and exposure.
- After enucleation the bony cavity is covered up by normal oral mucosa from edges of the lesion which grows in.

Limitations

The procedure requires a cooperative patient who will be available for follow-up regularly to get the cavity irrigated and packed with appropriate dressing. A longer follow-up is required because recurrence of keratocysts can take place over many years. Daughter cysts which theoretically are out pouching or budding of the cystic epithelium are expected to get reincorporated into the main cystic epithelium with this procedure. Still the fate of daughter cysts while performing the treatment modality is unknown.

Conclusion

Management of odontogenickeratocyst till date is a controversial subject in the field of oral and maxillofacial surgery with treatment modalities ranging from conservative to most radical. In young patients, looking at the functional and esthetic deformity produced as a result of radical treatment, many authors in different literatures have preferred conservative methods over radical methods in order to provide social, functional and psychological benefits to the patient. We in our institute have found this method of enucleation with open packing to be reliable and safe. This method is particularly useful where diagnosis of OKC is not confirmed on initial incisonal biopsy due to infective and inflammatory changes in the cystic lining. Sample size of our study is small (2 patients) and this method needs to be evaluated in more patients to make it as a viable treatment option for management of initially misdiagnosed OKCs.

REFERENCES

Betanieh AB. AL Qudah MA. 1998. Treatment of mandibular Odontogenic Keratocyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.,* 86:42-47

Brondum M, Jensen VJ. 1991. Recurrence of keratocyst and decompression treatment a long term follow up of 44 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.,* 72:265-269

Da Paula AN, Carvalhais JN, Domingues MG, Barreto DC, Mes Gupta RA. 2000. Cell proliferation markers in the odontogenickeratocyst, effect of inflammation. *J Oral Pathol Med.,* 29:477-482.

Eyre J, Zakrzewska JM. 1985. The conservative management of large odontogenickeratocyst. *Br J Oral Maxillofac Surg.,* 23:195-203

Maeda Y, Hiroto J, Yonedak K, Osaki T. 1990. Immunohisto chemical study of jaw cysts, different existence of keratins in odontogenic and non- odontogenic epithelial linings. *J Oral Pathol Med.,* 19:289-294

Nakamura N, Mitsuyasa T, Mitsuyasu Y, Taketomi T, Higuchi Y, Oshishi M. 2002. Marsupilization for odontogenickeratocyst: long term follow up analysis of the effects and changes in growth characteristics. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*., 94:543-553.

Ogden GR, *et al*. 1992. protein in odontogenic cysts: increased expression in some odontogenickeratocyst. *J ClinPathol.,* 45:1007-1010

Phillipson HP, 1956. OM Keratocyst (Kolestea – tomer I. Kaebeme, Tandiagebladet 60:963-980.

Williams TP, Connor FA. 1194. Surgical management of the Odontogenic Keratocyst: aggressive approach. *J Oral and Maxillofacial Surgery.,* 52:964-966

Marker P, Brondum N, Clausen PP, Bastion HL. 1996. Treatment of large odontogenickeratocyst by decompression and later cystectomy. *Oral Surg Oral Med Oral Pathol Oral RadiolEndod.,* 82:122-131.

Gorlin RJ, Golz RW. 1960. Multiple nevoid basal cell epithiloma jaw cyst. *N Engl J Med.,* 262:908-912

Pindborg JJ, Hansen J. 1963. Studies in odontogeniccystepithelium, clinical and roentgenologic aspects of odontogenic keratocyst. *AetaPthol Microbial Scand,* 58:283-294.

Shear M. 1960. Primordial Cyst. *J Dent Assoc S.,* 15:211-217

Browne RM. 1974. He Odontogenic Keratocyst histological features and their correlation with clinical behaviuor, *Br Dent J*., 131:249-259

Brannon RB. 1977. The OdontogenicKeratocyst a clinicopathological study of 312 cases. Part II Histological features. *Oral Surg Oral Med Oral Pathol Oral Radiol,* 43:233-255.

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