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RESEARCH ARTICLE

ODONTOGENIC KERATOCYST INVOLVING MANDIBLE-A CASE REPORT.

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Abstract

Odontogenic keratocyst (OKC) is a distinctive form of developmental odontogenic cyst. It has specific histopathologic features and aggressive clinical behavior including a high recurrence rate. OKC was first described by Phillipsen in 1956. OKC tends to grow in an antero-posterior direction within the medullary cavity of the bone without causing obvious bone expansion. This report describes a case on odontogenic keratocyst involving mandible occurring in a 25 years old patient.

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Introduction:-

The odontogenic keratocyst is a distinctive form of developmental odontogenic cyst that deserves special consideration because of its specific histopathologic features and clinical behavior. This cyst shows a different growth mechanism and biologic behavior from the more common Dentigerous cyst and Radicular cyst.¹

Jaw cysts constitute approximately 17% of intra-oral pathology.² The periapical cyst is the most common type of odontogenic cyst, followed by dentigerous and odontogenic keratocysts (OKCs).^{3,4} OKCs most often occur in the second and third decades of life and show a slight predilection for males (male to female ratio 1.3:1).⁴ The most recent WHO classification⁵ categorizes OKC as a developmental, noninflammatory odontogenic cyst that arises from the cell rests of the dental lamina. Harrington et al. best characterized this cyst by stating that "After thirty years of study, questions related to the histogenesis, pathogenesis, histology, high recurrence rate, and neoplastic potential of the OKC are still being debated."⁵

Odontogenic keratocysts may occur in any part of the upper and lower jaw with the majority occurring in the mandible, most commonly in the angle of the mandible and ramus.⁶ A localized asymptomatic swelling is the most common sign; spontaneous drainage of the cyst into the oral cavity and teeth mobility are also common. Radiographic appearances of OKCs are variable from a unilocular or a multilocular radiolucency with scalloped and well-defined margins to soap-bubble or honeycomb radiolucency.^{6,7}

The typical histologic features of the OKC include a thin, uniform layer of epithelium with little or no evidence of rete ridges, a well-defined basal cell layer with palisading cuboidal or columnar cells, and a corrugated, keratinizing luminal surface that is primarily parakeratinized. The presence of small "daughter" or "satellite" cysts may be observed in the connective tissue wall of the odontogenic keratocyst.⁸

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Meiselman et al consider conservative therapies to include “enucleation, curettage, and marsupialization.” Williams et al defines aggressive treatment as “that which is used in addition to enucleation, and includes curettage (mechanical, physical and/or chemical) and /or resection with or without loss of jaw continuity.”⁹

Here, we are presenting a case of 25 years old otherwise healthy male with swelling involving right anterior region of mandible which was diagnosed as odontogenic keratocyst based on clinical, radiological and histopathological features.

Case Report:-

A 25 years old male patient, from a semi urban area had reported to a private dental clinic with the chief complaint of a painless swelling involving right side of lower jaw since last 4 months without any history of discharge. The swelling was slow enlarging and asymptomatic. Initially, it was small in size, but had later grown gradually to attain the present dimension.

The past medical and dental histories were non-contributory. The patient was of average height and weight. Extra oral examination revealed a diffuse swelling involving right side of body of mandible (Fig. 1). On palpation, the swelling was non-tender. No regional lymph nodes were palpable. Intraoral examination showed the presence of a swelling along with buccal cortical plate expansion involving the alveolar ridge of mandible extending from the buccal aspect of 41 to 44 measuring about 3cm x 2 cm with obliteration of the labial vestibule (Fig. 2). There was no obvious swelling or lingual cortical plate expansion. On palpation, the swelling was fluctuant, non-tender. No paresthesia was noted. On aspiration straw colored fluid was obtained (Fig. 3).

PA view of face revealed an extensive radiolucent lesion involving the right body of mandible (Fig. 4). 30 degree lateral oblique view of mandible (right side) also revealed a large, multilocular radiolucent, osteolytic lesion involving the body of mandible, extending from 41 to 48 region. The lesion showed scalloped corticated margin (Fig. 5).

Considering the history, clinical and radiological findings, a provisional diagnosis of odontogenic keratocyst or ameloblastoma was made. Differential diagnoses included odontogenic myxoma, central giant cell granuloma, neurofibroma, central hemangioma etc.

Aspirated fluid analysis revealed Albumin : 3.3 mg/dl ; Globulin : 1.3 mg/dl ; Total protein : 4.6 mg/dl.

An incisional biopsy was performed under local anesthesia after routine blood evaluation (no significant alteration was noted) and informed consent from the patient, for histopathological evaluation.

Microscopic features of the H & E stained section showed the presence of cystic space lined by corrugated parakeratotic stratified squamous surface epithelium of 6-8 cell layers thick (Fig. 6). The basal epithelial layer was composed of a palisaded layer of tall columnar epithelial cell with hyperchromatic nuclei, arranged away from the basement membrane (Fig. 7). Flattening of the interface between the epithelium and connective tissue was also noted (Fig. 7). The underlying connective tissue was composed of fine collagen fibres and mild inflammatory infiltrate. No features of malignancy were seen. The overall histopathological features were corroborative to “**Odontogenic Keratocyst**”.

Depending upon the above diagnosis the patient was referred to the department of Oral and Maxillofacial Surgery for further management and treatment.

Discussion:-

Odontogenic keratocysts are generally thought to be derived from either the epithelial remnants of the tooth germ, or the basal cell layer of the surface epithelium.¹⁰ The majority of patients are in the age ranges of 20-29 years and 40-59 years. The distribution between sexes varies from equality to a male to female ratio of 1.6:1. Odontogenic keratocysts may occur in any part of the upper and lower jaw with the majority occurring in the mandible.¹¹ In the present case, odontogenic keratocyst was seen in a 25 years old male patient involving the anterior mandible, extending to the body.

OKC presents as asymptomatic swelling or associated with pain, trismus, sensory deficits, infection, drainage. Growth is chiefly in the anteroposterior dimension and the lesions may attain remarkable size without significantly deforming the jaw skeleton. This tendency to rapid growth is due to 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 higher accumulation of hyperkeratotic scales in the lumen of the cyst with resulting greater difference in hydrostatic pressure.¹² Here in this case, the patient reported with slow enlarging, painless swelling without any history of discharge.

Radiographically, it presents as a well-defined radiolucent lesion that is either unilocular or multilocular, with smooth and usually corticated margins, unless they have been secondarily infected.¹³ The present case revealed multilocular, extensive radiolucent, osteolytic lesion with scalloped corticated margin.

Biochemical analysis of Odontogenic keratocyst revealed the level of albumin 2-4 g/dl; globulin 0.5-2.5 g/dl and total protein <5g/dl.¹⁴ In the present case aspirated fluid analysis revealed Albumin : 3.3 mg/dl ; Globulin : 1.3 mg/dl ; Total protein : 4.6 mg/dl.

The histological diagnosis was based on the criteria as defined by the WHO, characterised by a thin fibrous capsule and a lining of keratinized stratified squamous epithelium, usually about five to eight cells in thickness and generally without rete pegs. Histologically OKCs have been classified into three categories: parakeratinised, Orthokeratinised, or a combination of the two types. Mostly (86.2%) were parakeratinised, 12.2% were Orthokeratinised, and 1.6% had features of both orthokeratin and parakeratin.¹⁵ In this case the cystic lining of the lesion was parakeratinized stratified squamous epithelium of uniform 6–8 cell thickness. The lining epithelium consisted of well-defined columnar basal cells in a palisade arrangement and with polarized nuclei. Special investigation like immunohistochemical analysis was not performed in this present case as the clinical, radiological, biochemical and histopathological findings were corroborative to the diagnosis of odontogenic keratocyst. Therefore, a final diagnosis of odontogenic keratocyst was made on the basis of clinical, radiological, biochemical and histopathological parameters.

Multiple treatments modalities for odontogenic keratocyst have been proposed and debated. There are proponents of “conservative” or “aggressive” methods of treatment.⁹ In our case, the patient was also advised for complete enucleation to reduce the post operative morbidity as patient is of very young age group and reported to the oral surgery department for further management and treatment.



Fig. 1:-Extraoral Photograph Of The Patient



Fig. 2:-Intraoral Photograph Showing A Swelling Involving Labial Mucosa



Fig. 3:-Straw Colored Aspirated Fluid



Fig 4



Fig 5

Fig. 4:-Pa View Of Face Showing An Extensive Radiolucent Lesion Involving The Right Body Of Mandible

Fig. 5:-30 Degree Lateral Oblique View Of Mandible(Right Side) Showing A Large, Multilocular radiolucent, Osteolytic Lesion Involving The Body Of Mandible, Extending From 41 To 48 Region With Scalloped Corticated Margin

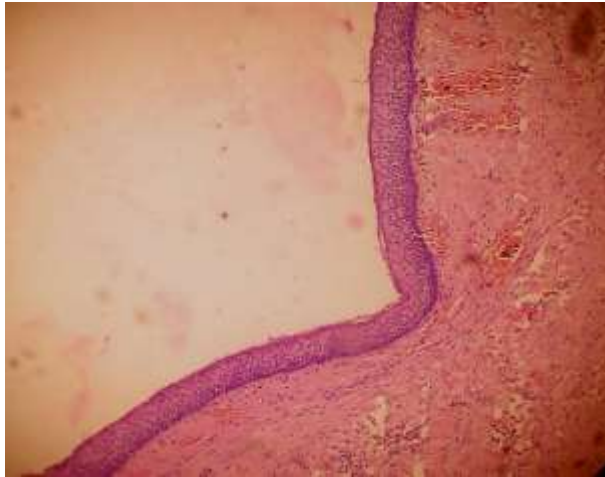


Fig. 6:-Low Power Photomicrograph Showing Presence Of Cystic Space Lined By Corrugated Parakeratotic Stratified Squamous Surface Epithelium Of 6-8 Cell Layers Thick

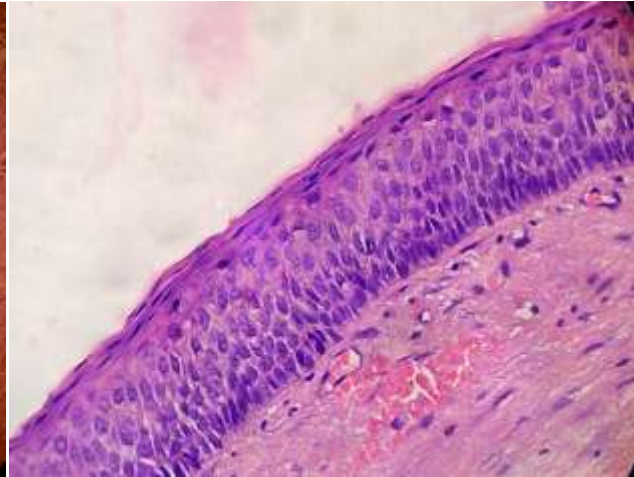


Fig. 7:-High Power Photomicrograph Showing Basal Epithelial Layer With A Palisaded Layer Of Tall Columnar Epithelial Cell And Hyperchromatic Nuclei, Arranged Away From The Basement Membrane With Flattening Of The Interface Between The Epithelium And Connective Tissue

Conclusion:-

The odontogenic keratocyst is a distinctive form of developmental odontogenic cyst that deserves special consideration because of its specific histopathologic features and clinical behavior. Careful clinical examination combined with thorough imaging modalities to evaluate the general aspects of the lesions and the margins, as well as its internal architecture and its relationship to adjacent anatomical structures are very important for proper diagnosis. These informations coupled with histopathological confirmation of the diagnosis will allow for the selection of the best individual therapeutic approaches, increasing the treatment efficacy in patients diagnosed with this odontogenic cyst. Thus, the clinicopathological, diagnostic and treatment modalities of odontogenic keratocyst, in general and a lesion involving the mandible, in particular is discussed herewith.

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