Journal Homepage: - www.journalijar.com
INTERNATIONAL JOURNAL OF ADVANCED RESEARCH (IJAR)
Article DOI: 10.21474/IJAR01/4430
DOI URL: http://dx.doi.org/10.21474/IJAR01/4430
ISSN NO. 2320-5407

RESEARCH ARTICLE

MASQUERADE OF A CYST: UNICYSTIC AMELOBLASTOMA.

Dr. Jochima Eudora Cota[1], Dr. Anita Spadigam[2] and Dr. Anita Dhupar[3].

1. MDS (Oral Pathology), Goa Dental College and Hospital.
2. Dean, Head and Professor, Department of Oral and Maxillofacial Pathology, Goa Dental College and Hospital.
3. Professor, Department of Oral and Maxillofacial Pathology, Goa Dental College and Hospital.

Manuscript Info

Manuscript History

Received: 09 April 2017
Final Accepted: 11 May 2017
Published: June 2017

Key words:-
Unicystic ameloblastoma, ameloblastoma, intraluminal, mural

Abstract

Unicystic ameloblastomas are a rare type of Ameloblastoma that clinically, radiographically present as a cyst but on histological examination show a typical ameloblastomatous epithelium. The concept of this tumour was first introduced by Robinson and Martinez in 1977. This is a case report of a 10 year old child presenting with a mandibular swelling and a peculiar radiological finding, highlighting the importance of histological examination in the diagnosis of unicystic ameloblastoma. A brief review of the pathogenesis of unicystic ameloblastoma and the various histological patterns that play a role in the diagnosis of a unicystic ameloblastoma and formulating the definitive treatment approaches for such lesions has been discussed.

Copy Right, IJAR, 2017,. All rights reserved.

Introduction:-

Unicystic ameloblastoma is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas (V. Nagalaxmi et al., 2013). This tumour was first introduced by Robinson and Martinez in 1977 (Yunus et al., 2009). This lesion shows clinical and radiographic characteristics of an odontogenic cyst but histologic examination is paramount in the diagnosis of the lesion (Gupta SS et al., 2011). This paper presents a case of a swelling of the mandible highlighting the same.

Case Report:-

A 10 year old boy presented with a slowly growing swelling on the right side of the mandible since one year. No history of trauma, pain, difficulty in swallowing or occlusion was reported. On physical examination face appeared asymmetrical with a diffuse swelling over the right side of the face. The skin was not attached to the underlying swelling and the inferior margin of mandible was continuous. Intraorally, a single, smooth swelling over the right mandibular alveolar ridge extending from 83 to 46 antero-posteriorly and causing expansion of the buccal cortical plate obliterating the buccal vestibule and slight expansion of the lingual cortical plate was noted. Overlying mucosa appeared blanched. Carious root piece of 83 was evident. On palpation, the swelling was bony hard, smooth and non-tender. No neck nodes were palpable. Systemic examination was normal. A provisional diagnosis of Keratocystic odontogenic tumour/OKC was agreed upon, with a differential diagnosis of dentigerous cyst, ameloblastoma and radicular cyst. An orthopantomogram (OPG) was done, which showed large unilocular radiolucent lesion in the right side of mandible associated with impacted 43, 44 and 45, 84 appeared to be within the cystic cavity. Mandibular true occlusal showed an expansion of the buccal cortical plate.

Corresponding Author:- Dr. Jochima Eudora Cota.
Address:- H.no. 42, Hapta, Betalbatim, Salcette, Goa, India, 403713.

On histological examination, a cystic lining with basal cells that appeared columnar to cuboidal with hyperchromatic nuclei showing reverse polarity (ameloblast like cells) were noted. Luminal proliferation was seen. The suprabasilar cells were loosely arranged resembling stellate reticulum like cells being consistent with a unicystic ameloblastoma.

Fig 1:-intraoral swelling

Fig 2:- OPG

Fig 3:- Mandibular True Occlusal

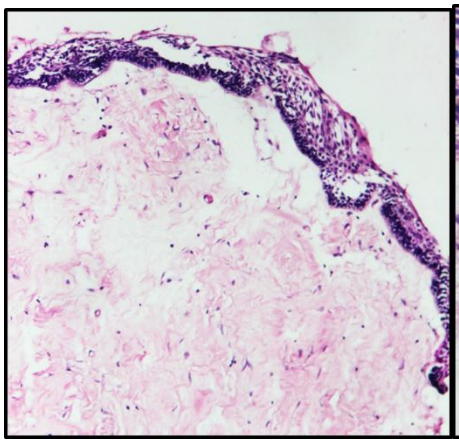

Fig 4:- H and E stained section (100x)

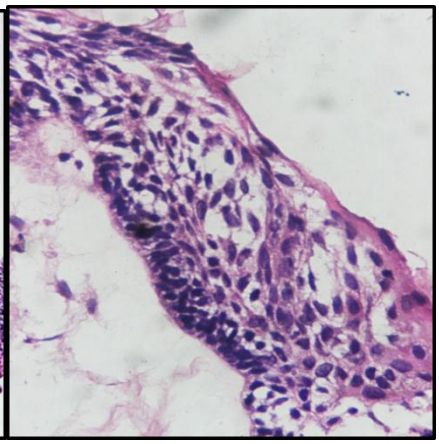

Fig 5:- H and E stained section (400X)

Discussion:-

Unicystic ameloblastoma usually occurs in younger age group and more than 90% are located in the mandible. (Figueiredo NR et al., 2015) Clinically it presents as a dentigerous type (associated with an impacted tooth) and a non-dentigerous type (not associated with teeth). (Deepalakshmi et al., 2017)

Leider et al. (Leider AS et al., 1985) proposed three pathogenic mechanisms for the evolution of UA: (1)The reduced enamel epithelium which is associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development. (2)Ameloblastomas arise in dentigerous cysts or in others in which the neoplastic

ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining. (3) A solid ameloblastoma undergoes cystic degeneration of the ameloblastic islands, with subsequent fusion of multiple microcysts and develops into unicystic lesions. The mechanism of cystic transformation is not certain, however the various theories include epithelial dysadhesion due to defective desmosomes or intrinsic production of proteinases like metalloproteinases, serine proteinases; the enzymes that normally degrade the central zone of the enamel organ after tooth development. (Rosenstein T., 2001)

Calretinin is a calcium binding protein found in normal human tissues and tumours like ameloblastoma. Studies have shown that calretinin is expressed only by UA indicating that it may be a specific marker for unicystic ameloblastoma (Anandani C et al., 2014; D'silva S et al., 2013). Bologna-Molina et al. (2008) found reduced expression of syndecan-1 in solid ameloblastoma when compared to unicystic ameloblastoma. Loss of syndecan-1 indicates unfavourable prognosis in epithelial tumours. Modolo et al. (2004) observed strong expression of the α1, α2, α3, α5, αv, β1, β3 and β4 integrins in luminal unicystic ameloblastoma. It was observed that a decrease in integrin expression is related to tumour growth and invasion of neighbouring structures. Various studies indicated that the proliferative markers like PCNA and Ki-67 were increased in the basal cells and cells of mural proliferations of unicystic ameloblastoma (Bologna-Molina R et al., 2013). High osteopontin expression and CD 44 v6 expression were found in unicystic ameloblastoma which is said to enhance tumour cell migration, invasion and spread. Twist is a mesoderm determining factor and it is a highly conserved basic helix loop transcription protein essential in embryological morphogenesis. Its high level in tumour will promote bone metastasis by bone remodelling. A higher expression is found in solid ameloblastoma as compared to unicystic ameloblastoma. (Zhong Y et al., 2011)

Ackermann classified this entity into the following three histologic groups:
Group I: Luminal UA, Group II: Intraluminal/plexiform UA and Group III: Mural UA
There is difficulty in determining the most appropriate form of treatment for these lesions. According to some authors, the presence of mural proliferation increases the rate of recurrence. For others, the choice of treatment for unicystic ameloblastoma, enucleation or surgical resection, depends on the severity and type of odontogenic epithelial mural proliferation (Garcia NG et al., 2016).

Philipsen and Reichart classified it as:

Subgroup 1: Luminal UA
Subgroup 1.2: Luminal and intraluminal
Subgroup 1.2.3: Luminal, intraluminal and intramural
Subgroup 1.3: Luminal and intramural

The first two groups of lesions may be treated successfully by enucleation or curettage; it has been suggested that recurrence following conservative surgery is more likely to occur in the third group and that these lesions should therefore be treated by radical resection, as for a solid or multicystic ameloblastoma (Garcia NG et al., 2016).

Marx and Stern classified the lesion as ameloblastoma in situ (developing in and limited to the epithelial lining of a cyst), micro-invasive ameloblastoma (arising from the epithelial lining and proliferating into the connective tissue layer of the cyst) and invasive ameloblastoma (arising from the epithelial lining and proliferation through the complete thickness of the connective tissue layer of a cyst). They suggested that ameloblastoma in situ and micro-invasive ameloblastoma should be treated with enucleation. Yet, invasive ameloblastoma should be treated with resection (Garcia NG et al., 2016).
The present case did not show any evidence of mural proliferation. Hence, considering the age of the patient and the histopathological features, a conservative mode of treatment in the form of enucleation was advised.

Conclusion:-

It is of utmost importance to correlate histopathologic findings with clinical and radiographic features to arrive at a correct definitive diagnosis. The Pathologist should examine the tissue sections carefully in an attempt to determine whether ameloblastoma has penetrated the wall of the cyst or not so that the complications can be minimized.

Bibliography:-

1. Alan S Leider, Lewis R Eversole, Michael E. Barkin. (1985). Cystic ameloblastoma: A clinicopathologic analysis. Oral Surgery Oral Medicine and Oral Pathology,, 6, 624–630.
2. Bologna-Molina R, Mosqueda-Taylor A, Lopez-Corella E, Almeida OP, Carrasco-Daza D, Garcia-Vazquez F, Farfan-Morales JE, Irigoyen-Camacho ME, Damián-Matsumura P. (2008, August). Syndecan-1 (CD138) and Ki-67 expression in different subtypes of ameloblastomas. Oral Oncology, 8, 805-11.
3. Chitra Anandani, Rashmi Metgud, Karanprakash Singh. (2014). Calretinin as a Diagnostic Adjunct for Ameloblastoma. Pathology Research International.
4. Modolo F, Martins MT, Loducca SV, de Araújo VC. (2004, September). Expression of integrin subunits alpha2, alpha3, alpha5, alphav, beta1, beta3 and beta4 in different histological types of ameloblastoma compared with dental germ, dental lamina and adult lining epithelium. Ora Disease, 10(5), 277-82.
5. Mohammad Yunus, Nahidulla Baig, Anwar Ul Haque, Adnan Aslam, Sundas Atique, Sobia Bostan, Adnan Mehdi Syed. (2009, June). Unicystic Ameloblastoma: A distinct Clinicopathologic Entity. Pakistan Oral & Dental Journal, 29(1), 9-12.
6. Natália Galvão Garcia, Denise Tostes Oliveira, Moacyr Tadeu Vicente Rodrigues. (2016). Unicystic Ameloblastoma with Mural Proliferation Managed by Conservative Treatment. Case Reports in Pathology.
7. Nigel R. Figueiredo, Manoj Meena, Ajit D. Dinkar, Sonam Malik, Manisha Khorate. (2015). Unicystic Ameloblastoma Presenting as a Multilocular Radiolucency in the Anterior Mandible: A Case Report. Journal of Dental Research, Dental Clinics, Dental Prospects, 9(3), 199-204.
8. Ronell Bologna-Molina, Adalberto Mosqueda-Taylor, Nelly Molina-Frechero, Ana D. Mori-Estevez, Guillermo Sánchez-Acuña. (2013). Comparison of the value of PCNA and Ki-67 as markers of cell proliferation in ameloblastic tumor. Medicina Oral, Patología Oral y Cirugía Bucal, 18(2), e174-e179.
9. S. Deepalakshmi1, Vasavi Santosh, Reema Manoj, Mandavi Waghmare. (2017). Cystic ameloblastoma in young girl of 18 years old: a non-dentigerous variant. Journal of Oral Medicine, Oral Surgery, Oral Pathology and Oral Radiology, 3(1), 73-76.
10. Sandeep S. Gupta, Shubhangi Mhaske, M.K. Gupta, Raju Ragavendra, Kavitha P Kamath. (2011, July). Unicystic Ameloblastoma Presenting as Multilocular Lesion: A Case Report. People's Journal of Scientific Research, 4(2), 55-57.
11. Shaloom D'silva, MK Sumathi, N Balaji, Nisha KN Shetty, KM Pramod, Jacob Cheeramelil. (2013, nov-Dec). Evaluation of Calretin Expression in Ameloblastoma and Non- neoplastic Odontogenic Cysts - An Immunohistochemical study. Journal of International Oral Health, 5(6), 42-8.
12. Tracy Rosenstein, M.Anthony Pogrel, R.A. Smith, Joseph A. Regezi. (2001). Cystic ameloblastoma—behavior and treatment of 21 cases. Journal of Oral and Maxillofacial Surgery, 59(11), 1311–1316.
13. V. Nagalaxmi, Mithare Sangmesh, Kotya Naik Maloth, Srikanth Kodangal, Vani Chappidi, Stuti Goyal. (2013). Unicystic Mural Ameloblastoma: An Unusual Case Report. Case Reports in Dentistry.
14. Yi Zhong, Wei Guo, Li Wang, Xinming Chen. (2011). Molecular markers of tumor invasiveness in ameloblastoma: An update. Annals of Maxillofacial Surgery, 1(2), 145-149.