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RESEARCH ARTICLE

CLINICAL AND RADIOGRAPHIC DILEMMA: A CASE OF ADENOMATOID ODONTOGENIC TUMOR OF MANDIBLE

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Abstract

Adenomatoid Odontogenic Tumor (AOT) is benign epithelial lesion of odontogenic origin derived from complex system of dental lamina remnants. The lesion is well known for various clinical and histopathological appearances and so it is has been given the title 'master of disguise'. It is well-recognised as a leisurely growing benign tumor which is not rare now as it was previously reported. This lesion is grouped into three variants of which the most common variant is follicular type which often presents as dentigerous cyst clinically because it surrounds an impacted tooth. We report an AOT of mandible in a 14-year-old male who reported to our department and was provisionally diagnosed as unicystic ameloblastoma or dentigerous cyst. The article also covers a review of literature on the tumor. Clinical, radiological, histopathological characteristics and treatment modality of the case have been stressed upon.

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Introduction:-

The lesion now known as the adenomatoid odontogenic tumor, was documented as a separate entity by Stafne^[1]. The earliest recorded case was in 1907 by Driebladt who described it as a pseudo-adenoma- adamantinoma. This tumor was assigned various names until Philipsen and Birn introduced the name 'Adenomatoid odontogenic tumor' in 1969. The histologic typing of WHO in 1971 introduced a definition for this lesion describing it as, "A tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The lesion may be partly cystic and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm"^[2]. In 2005 a new definition was proposed as, 'A tumor composed of odontogenic epithelium, presenting a variety of histoarchitectural patterns, embedded in mature connective tissue stroma and characterized by slow and progressive growth.' It is now a not so rare benign epithelial lesion derived from the remnants of dental lamina enveloping a developing dental follicle which is mostly solid and may be partly cystic. AOT constitutes 2.7 to 7% of all odontogenic tumors^[3]. It is primarily found in young females with a male:female ratio of 1:2, occurring largely in the maxilla in association with an impacted permanent tooth. For radiological diagnosis, the intraoral periapical radiograph holds more diagnostic value than panoramic. AOT exhibits resemblance to other odontogenic unilocular cystic lesions enclosing the unerupted teeth such as dentigerous cyst or ameloblastoma in its clinical and radiographic appearance. Histopathologically also, the lesion is sometimes found to contain a cystic component. Based on the clinical and radiologic findings, AOT can be subdivided into two variants: (A) Central (or intraosseous) variants 1) Follicular type which encompasses the crown of an unerupted tooth thereby, resembling a dentigerous cyst. 2) Extrafollicular type has no association with the crown of an unerupted tooth. The provisional diagnosis of this variant could be a 'residual', a "globulomaxillary" or a lateral periodontal cyst depending on the actual intraosseous localization of the lesion. B) Peripheral (or

extraosseous) variant with a resemblance to gingival fibroma or fibrous epulis^[4].

Case Report:

A 14 year old male patient reported to the department of Oral and maxillofacial surgery of Saraswati Dental College with the chief complaint of swelling in the left lower front region of jaw for 4 - 5 months. The swelling gradually increased to its present size. On examination it was measured to be about 5x4 cm. Mild mobility in teeth 31,32,73,34,35 was observed in the teeth associated with the lesion. Deciduous canine was retained. The swelling was ovoid in shape obliterating the buccal vestibule along with expansion of both cortices(Figure 1). There was no paresthesia in the distribution of Mental nerve. On radiographic examination a mixed radio-opaque - radiolucent lesion was seen on the left mandibular region(extending from symphysis to body) with multiple areas of cortical perforations. The permanent canine was enclosed within the confines of tumor mass approximating the lower border of mandible (Figure 2) However, the patient was asymptomatic. The aspiration was negative.

Expansion of both the cortices was evident in the CT scan(Figure 3) . Incisional biopsy was done after routine blood investigations and the histopathological report confirmed the lesion to be adenomatoid odontogenic tumor, follicular type. Pre- anesthetic evaluation of the patient was done and arch bar was placed in both the jaws. The surgery was performed under general anesthesia under complete aseptic conditions. A crevicular incision was made from the left central incisor to 2nd premolar region in the buccal vestibule and mucoperiosteal flap was raised. The buccal cortical plate was entirely resorbed, interspersed with thin friable bone. The lining of the tumor mass was completely excavated from the bony cavity along with the associated teeth from central incisor to 2nd premolar(Figure 4,5,6). The wound was copiously irrigated with betadine and saline.

Iodoform dressing was placed within the cavity formed to promote secondary healing. Precautionary post operative IMF was done. The dressing was changed every 3 days in the beginning for 15 days, weekly later on and now it is changed in every 15 days. The healing has been observed to be good with no post-operative complications (Figure 7,8,9) and new bone formation can also be appreciated. 1 year post-operative picture shows complete healing.

The resected tumor mass histopathologically showed features of adenomatoid odontogenic tumor comprising sheets/ nests/ whorled masses of columnar/cuboidal cells. Foci of calcification were also seen. Eosinophilic, uncalcified, amorphous material was found, suggestive of "tumor droplets" which is one of the characteristic feature of AOT.

Discussion:-

AOT is an imitator tumor mimicking Dentigerous cyst and Unicystic ameloblastoma very frequently. Therefore, the origin of this tumor has also been a topic of debate as there are two schools of thought describing the origin of this tumor. According to Philipsen and Reichart, the dental lamina remnants could probably be the parent cells for this tumor. This theory states that the lesion grows adjacent to or into a dental follicle constituting the "envelopmental theory"^[7,8]. Santos et al.^[9,10] and Garcia Pola et al.^[10,11] reported cases of this tumor originating from the odontogenic epithelium of a dentigerous cyst. Cassiano Francisco Weege et al.^[7] also mentioned such a case. This tumor is documented to involve not only the anterior maxilla but other regions of the jaw such as the angle of the mandible. The reason for this correlation between an odontogenic cyst and tumor can be attributed to the fact that neoplastic and hamartomatous changes can begin at any stage of odontogenesis. This necessitates taking biopsy sample from two sites. Iron-binding proteins and proteinase inhibitor may be involved in the pathogenesis of AOT.^[6]

Various studies and literature support the fact that the maxillary arch is the predominant site of occurrence, being almost twice as frequent as that of the mandible. Giansanti et al. (1970) reported that 65% AOTs were seen in the maxilla and 35% in the mandible. In a recent retrospective study of 61 cases in Nigerian population, 55.8% cases were seen in maxilla whereas 32.8% cases were found in the anterior mandible^[12]. However, this study was conducted on a small population. Another study which was conducted in 2007 by Philipsen and Reichart analysed 1082 cases on the basis of clinical and epidemiological profile found 64.3% cases in maxilla and 35.7% cases affecting mandible similar to other studies. This review was based on the largest number of cases ever presented. Chandramani B More found that of the lesions found in the mandible, 69% were present in the anterior region, 27% in the premolar region, and a few in the molar region^[13]. Mandibular AOTs are similar to maxilla in their clinical as well as radiographic pattern. Though, mandibular AOTs are a rare finding one should never ignore the possibility of finding it in the mandibular anterior region when there is an impacted tooth associated with it.

Epithelial tumor cell components in AOT have been described into three cell types by Takahashi et. al. Cell type I: small compact cells in a solid nodule and pseudoglandular cells in a duct-like structure; Cell type II: peripheral elongated cells and spindle shaped cells in a cribriform pattern; and Cell type III: metaplastic squamous cells^[6]. Calcifying epithelial odontogenic tumour (CEOT)-like areas are also found in some cases of AOT.

Immunohistochemically AOT cases show positive results for AE1/AE3, 34bE12, CK5, CK14 and CK1. Vimentin is also expressed in some cases and this may suggest the pattern of varied phenotypical characteristics in certain areas of the tumor.

Recurrence of this tumor has been reported to be very low. The low recurrence after surgical treatment can be attributed to the low proliferative activity observed in cases with Ki-67 marker³. In a survey of 21 cases from Srilanka by Mendis et al in 1990, no recurrence was observed after conservative surgery in a follow up period of 2-11 years. In a study conducted in 2005 by Leon et al. also there was no recurrence found after surgical treatment. Only three cases in Japanese patients are reported in which the recurrence of this tumor occurred^[16]. One of these cases was reported to have intracranial extension from the maxilla by Takigami and Ueda et al in 1988.

These lesions can grow to considerable sizes also involving the inferior border of the mandible. Owing to their benign nature, slow growth and clear margins, as well as low propensity to recur, the treatment followed till date is enucleation and simple curettage, although in exceptional cases of large tumors, there is risk of bone fracture. Partial resection, en bloc of the mandible or maxilla has been indicated for such cases^[14]. Additionally, lyophilized bone and guided tissue regeneration find use in cases where surgical enucleation leave large exposed osseous cavities^[14]. Due to its imitating nature, iatrogenic resection of body of mandible can also be performed. The resection of mandible in young patients still in their growing years is associated with plethora of complications such as loss of jaw bone support, deformity, dysfunction and psychological trauma even after good reconstruction. Therefore, an alternative conservative surgical procedure “Dredging Method” which has been reported with good success rate in ameloblastoma by Sma Sadat and M Ahmed^[15], can also be tried for large, aggressive AOTs. This calls for more research in this area.

Studies conducted on the use of iodoform dressings in OKC found very low recurrence in these tumors when they were managed conservatively, (Giuliani, 2006, Zhou,2012) followed by iodoform dressings postoperatively.^[17] Iodoform dressing was, therefore used in our case as it is an antiseptic which facilitates healing of the tissue in which it is placed by providing an aseptic environment. The histopathology report revealed typical features of adenomatoid odontogenic tumor, follicular type which usually establishes itself around the crown of unerupted, anterior teeth in growing patients, being constituted of whorled nests of duct like epithelial cells together with areas of glandular or ductal patterns intermixed with occasional spherical calcifications.

It is interesting to note that irrespective of the pattern, the biologic behavior of the tumor never changed unlike that of the other tumors as confirmed by various studies regarding the same. Although it presents with varied histopathological patterns, its histology also has always remained distinct making it easy to diagnose this tumor. This requires attention for emphasizing more on the histomorphology of AOT and in deriving why none of these patterns affect the biological behaviour of these tumors.

Conclusion:-

It should be emphasized that AOT is very rare in mandible and when present it is most commonly diagnosed as a dentigerous cyst or unicystic ameloblastoma leading to wrong diagnosis. It is therefore mandatory to wait for biopsy report before proceeding for any treatment and it should also be stressed that the biopsy sample should be taken from two different sites of the tumor to rule out the possibilities of any other lesion, like dentigerous cyst or CEOT. Once confirmed with the diagnosis, conservative treatment should be employed as the tumor has a good prognosis and young individuals have good healing potential.

Figure legends



Figure 1

Preoperative clinical



Figure 2

OPG demonstrating the lesion

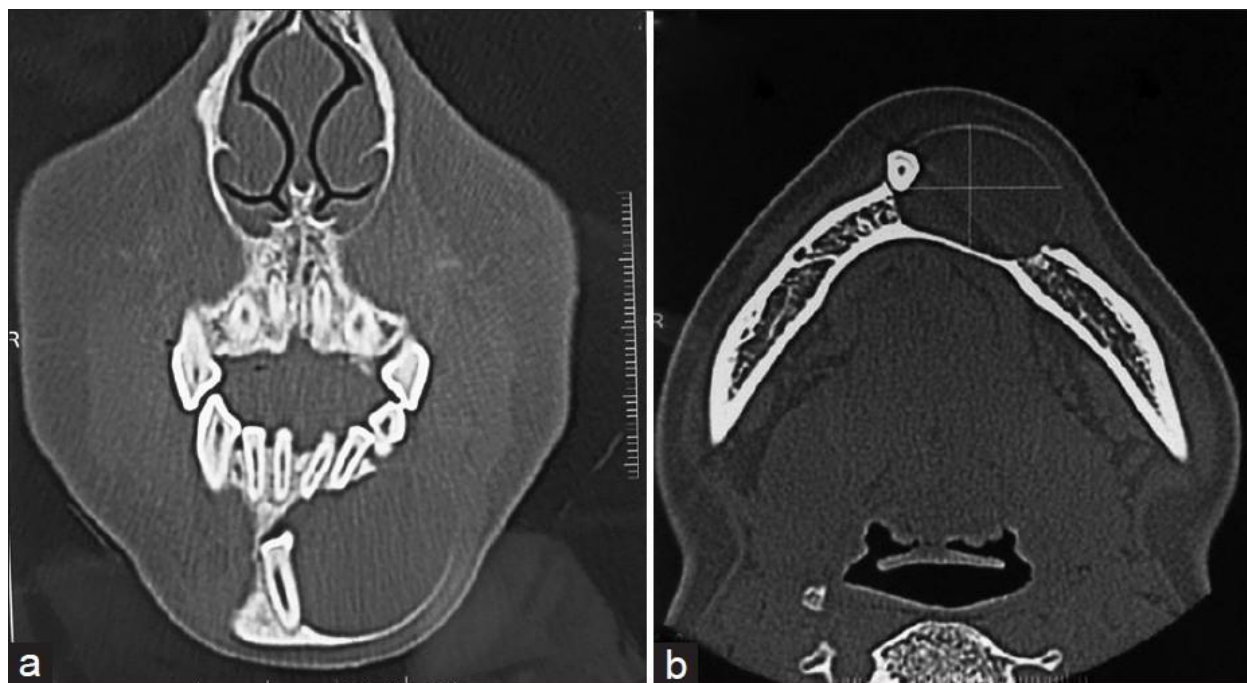


Figure 3

CT scan demonstrating the tumor



Figure 4



Figure 5

Intraoperative images demonstrating resection of tumor mass along with the resected tumor.



Figure 6

Gross specimen of resected tumor



Figure 7

3 months post-op image demonstrating good healing.



Figure 8

3 months post-op OPG with iodoform dressing in place depicting the reduced size of the cavity and formation of new bone.



Figure 9

1 year post-op

Ethical approval:

Written informed consent from the patient's guardian was obtained.

Conflict of interest:

There is no conflict of interest regarding this publication.

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