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RESEARCH ARTICLE

COMPLEX ODONTOME: A RARE CASE REPORT WITH LITERATURE REVIEW

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Abstract

Odontomas are the most common variety of odontogenic tumors and derived from differentiated epithelial and mesenchymal cells. Traditionally, odontomas have been classified as benign odontogenic tumors and are subdivided into complex or compound odontomas morphologically. This paper describes the case of a complex odontoma in a 10-year-old girl diagnosed after extraction. A surgical excision was performed and the histopathological examination revealed a complex odontome. They usually cause delayed eruption, so early detection and treatment of odontomas could decrease the possibility of late eruption or even tooth impaction due to overlying odontomas.

Introduction:-

The term "odontoma" was first coined by Paul Broca in 1866, who defined the term as tumor formed by the overgrowth of complete dental tissue.[1] They are the most common benign odontogenic tumor of epithelial and mesenchymal origin.[2] The growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts results in the formation of these tumor. These cells result in formation of variable amounts of enamel, dentin and pulpal tissue of the odontoma.[3] Enamel and dentin are usually laid down in aberrant pattern since the organization of odontogenic cells fail to reach the normal state of morphodifferentiation.[4] So, are considered as developmental anomalies rather than true neoplasm. Odontomas majorly comprises enamel and dentine but variable amount of cementum and pulp tissue can also be found.[5] Odontoma can be broadly classified into compound and complex odontoma. When the deposition of enamel and dentine during the development of tumor is in such a way that the resulting structure show an anatomic similarity to normal teeth, the lesion is classified as a compound odontoma. Whereas, in complex odontoma there is deposition of simple irregular mass occurring in irregular pattern.Compound odontoma is more common than complex odontoma.[7]

The incidence of compound odontoma ranges between 9 and 37% and the complex odontoma is between 5 and 30%.[8] The majority of odontomas of anterior segment of the jaws are compound composite type (61%), whereas the majority of posterior segment are complex composite type (34%). Both type of odontomas occur more commonly on the right side than on the left, (compound 62%, complex 68%).[4] The compound composite odontoma most frequently occur in incisor cuspid region of the upper jaw in contrast to the complex odontoma which are commonly found in molar and premolar region of the mandible.[9] However, these tumors can be found anywhere in the dental arch but have less association with primary dentition.

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The odontoma has unknown etiology. However, it has been suggested that the ideal conditions for its appearance is offered by trauma and infection at the place of the lesion.[4] Odontoma are generally asymptomatic, have slow growth, and rarely exceed size of a tooth, but can expand the cortical bone when size is large.[5]

Odontomas are usually detected during the first two decades of life, but they may be diagnosed at any point of time.[5] Analysis of 396 cases in a study showed that the diagnosis usually happens between 11 and 15 years of age. Odontomas are frequently associated with unerupted teeth leading to its impaction. The most frequently impacted teeth by odontomas are the canines, followed by upper central incisors and third molars.[4] Generally these tumor remains inside the bone (intraosseous), but occasionally they may erupt into the oral cavity.[10]

Odontomas have characteristic radiographic aspects. An irregular mass of calcified material surrounded by a thin radiolucent area with smooth periphery is suggestive of complex odontoma. The compound type shows calcified structures resembling teeth in the centre of a well-defined radiolucent lesion. A developing odontoma can be discovered by routine radiography, but due to lack of calcification, it may cause difficulty in identification.[4] The histological examination of odontomas often shows the presence of enamel matrix, dentin, pulp tissue, and cementum that can, but need not, exhibit a normal relationship. Compound odontomas are formed by tooth-like structures which resemble pulp tissue in the central portion surrounded by a dentine shell and partially covered by enamel. Complex odontomas are conglomerates without orientation of dentin, enamel, enamel matrix, cementum, and areas of pulp tissue. The capsule of connective tissue surrounding an odontoma is similar to the follicle that covers a normal tooth.[4]

Treatment of odontomas include conservative surgical removal with very less probability of recurrence.[5] A great radiographic resemblance is seen between ameloblasticfibroodontomas and odontoameloblastomas to common odontomas. Therefore, all specimens should be sent for histopathological examination.[10] Besides, proper care of the patient should include cautious clinical and radiographical follow-up.[8]

Table 1:- Major Characteristics of Compound and Complex Odontomas[9].

Major Characteristics	Compound Odontoma	Complex Odontoma
Frequency	The relative frequency among odontogenic tumors varies between 9% and 37%. It's considered the commonest odontogenic malformation.	The relative frequency among odontogenic tumors varies between 5% and 30%.
Age	The majority of cases appear before the age of 20, making it a lesion of childhood/adolescence.	The majority of cases occur before the age of 30 with a peak in the second decade of life.
Gender	Male and female subjects are equally affected.	Male and female subjects are equally affected.
Sites	Maxillary anterior region is the most frequent site.	Posterior mandibular followed by anterior maxilla are the most frequent sites.
Clinical presentation	Painless, non-aggressive lesion, with a more limited potential growth than the complex odontoma. Often associated with an unerupted permanent tooth.	Painless, slow-growing and expanding lesion. Often associated with an unerupted permanent tooth.
Radiological features	Radiopaque mass of multiple, small, calcified structures with an anatomical similarity to normal teeth usually surrounded by a narrow radiolucent zone.	More or less amorphous mass of calcified material with the radiodensity of tooth structure, which bears no anatomical resemblance to tooth, surrounded by a narrow radiolucent rim
Treatment	Conservative surgical enucleation	Conservative surgical enucleation.

Case Report:

A 10-year-old girl presented to the department of Paediatric and Preventive Dentistry, Subharti Dental College & Hospital, Meerut (U.P.) with the chief complaint of rotated tooth in the lower right front tooth region (Fig. 1A). A digital orthopantomogram (Fig. 1B) was advised as a routine dental investigation for orthodontic treatment. An irregular mass with respect to the root of the tooth which was rotated was observed. Provisional diagnosis of

odontoma was made which was confirmed by histopathological examination after resection. On cone beam computed tomography (CBCT) examination, to find the extent of the mass, it was found that the odontoma was attached to the root and was in lingual respect to the tooth. The mass was approximately 9.5 mm supero-inferiorly, 10.5 mm buccolingually and 7.5 mm mesio-distally (Fig. 1C). Surgical resection of the mass was planned with extraction of 41.

Complete blood count, bleeding time and clotting time was advised prior to the surgery and informed consent was taken. Scaling was done prior to start the treatment (Fig. 2A) and patient was on antibiotics from a day prior to the surgery. After part preparation, bilateral inferior alveolar nerve block was administered and full thickness mucoperiosteal flap (trapezoidal) was raised with two vertical incision on mandibular right canine and mandibular left lateral incisor for better access to the surgical site. Buccal cortical plate over the odontoma was expanded which served as a guide for bone guttering. Bone guttering was done over the raised buccal cortical plate and the odontoma was seen. Calcified structure together with the permanent mandibular right central incisor was resected. The surgical site was irrigated with saline followed by betadine and bleeding was induced. Suturing was done using 3-0 silk suture for 1 week (Fig. 2B). Immediate post-operative intra oral peri apical radiograph was taken to ensure the complete resection of odontoma (Fig. 2C). The resected sample (Fig. 2D) was then sent for histopathological examination to the department of oral and maxillofacial pathology and oral microbiology.

After 1 week the sutures were removed and the surgical site was again irrigated with betadine (Fig. 3A). One-week follow-up IOPA radiograph was taken (Fig. 2C). The histopathological report showed disordered mixture of dental tissues composed of cementum-like areas admixed with dentin with dentinal tubules and small areas of pulp tissue inclusions. Empty spaces suggestive of enamel spaces are also seen. Cellular connective tissue stroma suggestive of ecto-mesenchyme with an island of odontogenic epithelium is seen at one corner of the section (Fig. 2B). These features were suggestive of complex odontoma.

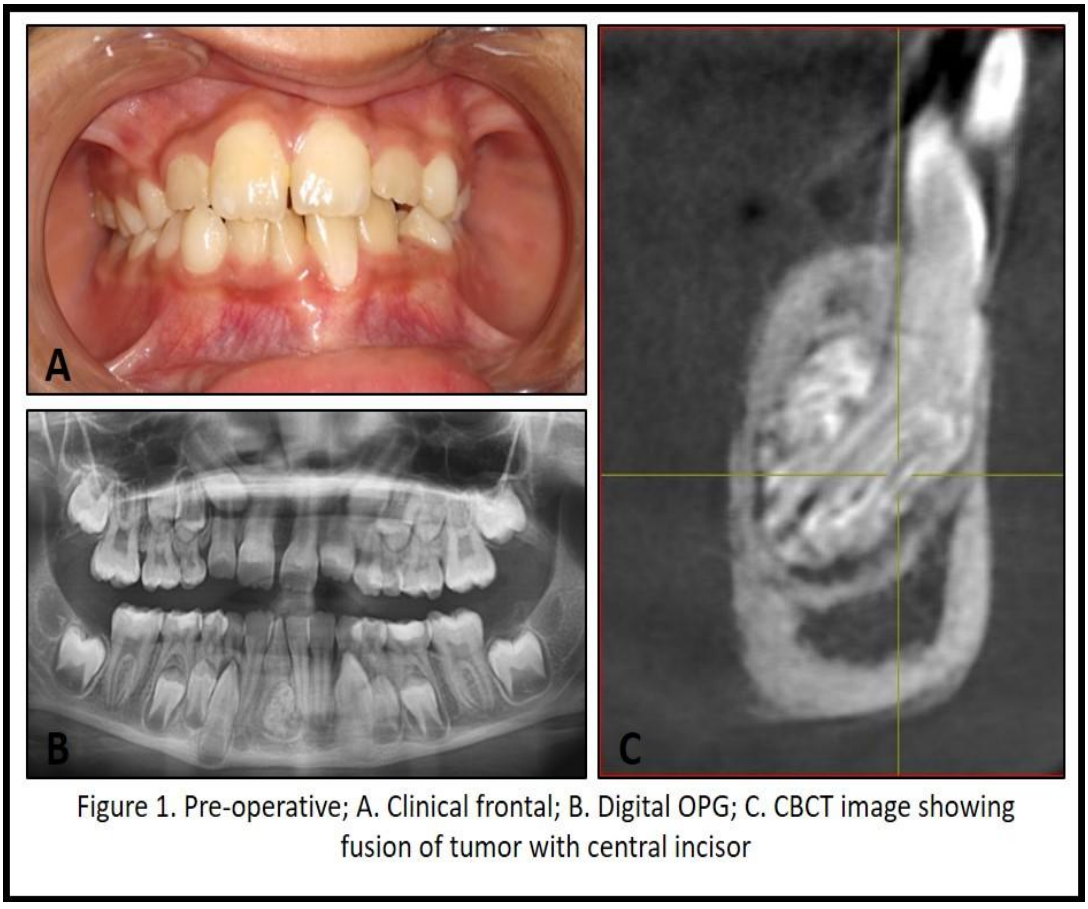

Figure 1. Pre-operative; A. Clinical frontal; B. Digital OPG; C. CBCT image showing fusion of tumor with central incisor

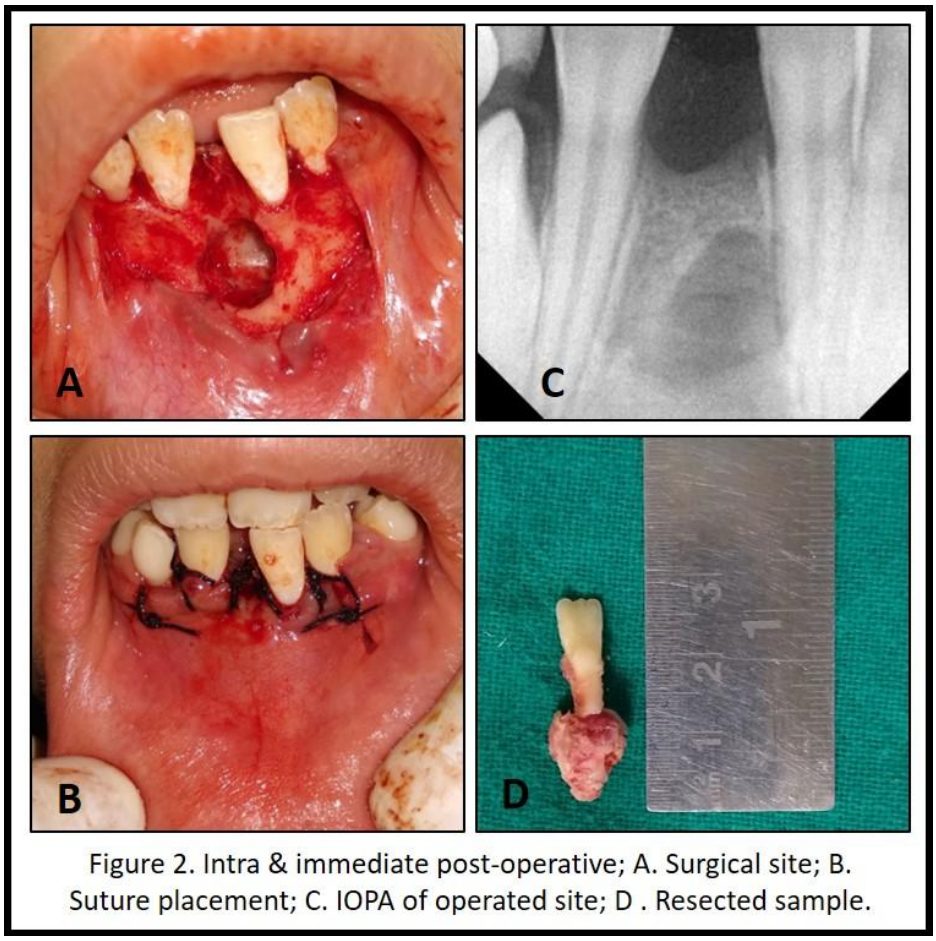

Figure 2. Intra & immediate post-operative; A. Surgical site; B. Suture placement; C. IOPA of operated site; D . Resected sample.

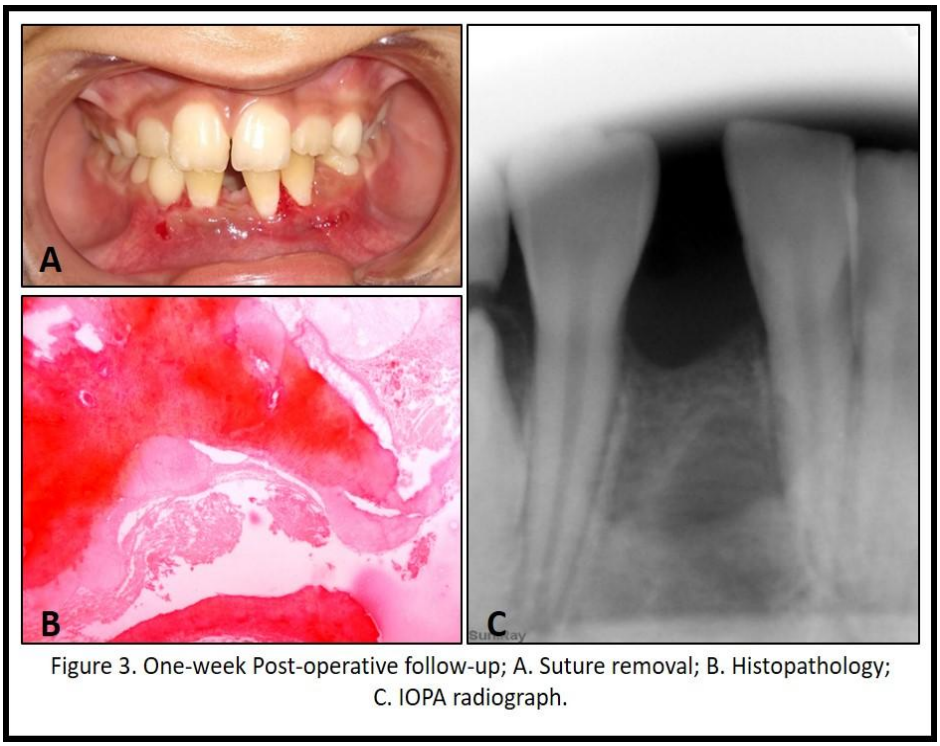

Figure 3. One-week Post-operative follow-up; A. Suture removal; B. Histopathology; C. IOPA radiograph.

Discussion:-

Odontoma are benign tumor of the jaw that consist of enamel, dentine, cementum and pulpal tissue.[11] They are relatively common odontogenic lesions, generally asymptomatic, and are rarely diagnosed before the second decade of life. In different reports and populations, the frequency of occurrence of odontoma varies greatly. In Caucasian population, odontomas are reported as the most frequent odontogenic tumor by Sriram and Ravindra[12]. They also reported 6% incidence of odontomas in odontogenic tumor. Buchner et al.,[13] reported incidence of 75.9% odontoma among all odontogenic tumors. A controversial data exists regarding gender predilection. According to some authors, greater incidence was reported among females,[13] while others claims among males[4]. Whereas, the third group of studies reports showed no difference between males and females.[11] Impaction or delayed eruption of permanent teeth can be frequently caused by odontoma.[5]

This paper describes two cases of odontoma, one of each variant i.e. compound and complex. This diagnosis was later confirmed by histological examination of the lesions after their surgical resection. Complex and compound odontoma is most commonly found in posterior mandible and maxillary anterior region respectively. Alternatively, this paper reports a complex odontoma in mandibular anterior region and a compound odontoma in mandibular posterior region, which makes them unique.

There are several classifications of odontoma, few of them are as follows:

WHO Classification:

1. One of the most common classifications is given by World Health Organization (WHO). Four lesions containing enamel and dentine of normal appearance are defined in the WHO classification.[14] They are as follows:
2. Ameloblastic fibro-odontome: Consists of varying amounts of calcified dental tissue and dental papilla like tissue, the latter component resembling fibroma. The ameloblastic fibro-odontome is considered as an immature precursor of complex odontome
3. Odonto-ameloblastoma: It's a very rare neoplasm which resembles an ameloblastoma both structurally and clinically but contains enamel and dentine
4. Complex odontome: When the calcified dental tissues are simply arranged in an irregular mass bearing no morphologic similarity to rudimentary teeth
5. Compound odontome: Composed of all odontogenic tissues in an orderly pattern that results in many teethlike structures but without morphologic resemblance to normal teeth.

On the basis of gross, radiographic and microscopic features[1], two types of odontoma are recognized, namely:

1. Compound and
2. Complex.

On the basis of their developmental origin[2], it can be grouped into three types:

1. Epithelial
2. Composite (epithelial and mesodermal) and
3. Connective tissue.

According to their position within the jaws:[15]

1. Intraosseous (erupted odontoma): They occur inside the bone and may erupt into the oral cavity.
2. Extraosseous or peripheral odontomas: These are odontomas occurring in the soft tissue covering the tooth bearing portions of the jaws, having a tendency to exfoliate.

Thoma and Goldman (1946) classification:[16]

1. Germinated composite odontomes—two or more, more or less well-developed teeth fused together
2. Compound composite odontomes—made up of more or less rudimentary teeth
3. Complex composite odontomes—calcified structure, which bears no great resemblance to the normal anatomical arrangement of dental tissues
4. Dilated odontomes—the crown or root part of tooth shows marked enlargement
5. Cystic odontomes—an odontome that is normally encapsulated by fibrous connective.

Z Gorlin et al. eliminated the term composite as redundant and classified odontomas as either complex or compound.

There are essentially two types of odontome:[17]
1. Complex composite odontome
2. Compound composite odontome.

A new type known as hybrid odontome is also reported by some authors.

According to Robinson,[18] in 1952, in his classification restricted the term odontome for those tumors which aroused from both epithelial and mesenchymal dental forming tissues. But presently, this term is used in a very restricted sense to designate only those tumors which consist of dental hard tissues.

Compound Odontome:
These are the malformations in which all dental tissues are represented in a more orderly pattern, so that the lesion consists of several tooth-like structures or denticles composed of enamel, dentin, cementum and pulp. It is a tumor of enamel and dentin arranged in the form of anomalous miniature teeth. Several small abnormal teeth might be surrounded by a fibrous sac.

Complex Odontome:
These are the malformation in which all dental tissues are represented but not in an organized form or disorderly pattern. It is an odontogenic tumor characterized by the formation of calcified enamel and dentin in an abnormal arrangement because of lack of morphodifferentiation.

In case of observation of a radiographic finding resembling odontomas, differential diagnosis must be established with several lesions depending on its location. If the lesion occurs at inter-root location, then cementoma, focal residual osteitis, calcifying epithelial odontogenic tumors (CEOT), adenomatoid odontogenic tumors (AOT), supernumerary teeth or benign osteoblastoma should be included in the differential diagnosis. If the lesion is located at pericoronal level, then AOT, CEOT should be included in the differential diagnosis. When the lesion occurs in the maxillary sinus, then it could be misdiagnosed with antral mycosis, sinusitis, foreign bodies, retained root, benign mesenchymal neoplasms, peripheral osteoma, antral sarcoma or carcinoma. It can also be associated with some syndromes such as basal cell nevus syndrome and Gardner syndrome.[19] It is suggested that there should be better routine clinical and radiological follow-up, to avoid carcinoma, adenomatoid tumor or ameloblastoma.[20]

In our case, the chief complaint of the patient was irregular teeth in lower front tooth region of the jaw. There was no history of pain and extraoral swelling. Odontoma was diagnosed in routine OPG with respect to lower right central incisor. CBCT and histopathological findings suggested the presence of complex odontoma in lingual relation to the mandibular right central incisor. The odontoma was fused with the cementum of the tooth so, the treatment included extraction of the offending tooth. The diagnosis was made during the period of primary mixed dentition.

Conclusion:-
Odontomas are one of the benign tumors that are frequently seen in both jaws. They have been detected in routine radiographic examination only if they are asymptomatic. They usually cause delayed eruption, so early detection and treatment of odontomas could decrease the possibility of late eruption or even tooth impaction due to overlying odontomas.

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