Adenomatoid Odontogenic Tumour of the Maxillary Sinus

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Abstract

A typical adenomatoid odontogenic tumour removed from the left maxillary sinus of a fifteen years old male is reported

The adenomatoid odontogenic tumour or adenoblastoma is a distinct tumour of odontogenic origin. This lesion has been described as early as 1907, but Stafne delectated it as separate entity in 1948, for the first time.

It is a benign lesion which arises from the residual odontogenic epithelium, most frequently in the second decade of life. Females are affected in nearly two- third of cases , maxilla is involved almost twice as often as the mandible. In 76% of cases the anterior part of the maxilla is involved. Most of the tumours (74%) are associated with unerupted teeth , the canines are affected 68% of the time. A high association with odontogenic cyst is noted.

The clinical and pathological features with review of literature of this uncommon lesion is presented here.

Case Report

A Fifteen years old male reported in the ENT OPD with complaints of swelling on the left side of the face in the maxillary region since one year. (X.12 x2) Swelling was gradually increasing in size, hard to touch, smooth, nontender extending all over maxilla. Dentition was normal. Skin over the swelling was normal and there was no other positive findings. Patient was healthy, well built and well nourished. Nasal patency on left side was reduced due to pushing of the lateral wall towards the septum. No proptosis seen. Left infraorbital margin was intact. X-ray PNS showed a homogenous, well demarcated circumscribed area of opacity with expansion and no bony erosion in left maxillary sinus. No unerupted tooth in the sinus was seen (Fig.1). Provisional diagnosis of odontogenic tumour was made. Patient was operated through sublabial approach, Caldwell-Luc was done. A large cystic mass of 7.5 x 3.5 cm size was taken out completely containing lots of fluid and a small tissue attached to medial wall of the cyst (mural tissue) (Fig.2)

Fig.1. Photo face of the Patient

Fig. 2. Gross specimen of cyst showing well defined capsule and mural excrescences.

Microscopically the tumour is well encapsulated and composed of two type of structures : duct like structures lined by single layer of columnar epithelium cells and groups of polyhedral and spindle shaped cells showing whorling pattern. The nuclei of the duct cells are arranged regularly. The ducts contain eosinophilic hyalinised material. No pleomorphism or mitosis are seen in these cells. Histopathological examination revealed Adenomatoid odontogenic tumour. Postoperative period was uneventful and the patient was healthy (Fig. 3).

Review of Literature

The odontogenic adenomatoid tumour is a rare odontogenic lesion that has been reported under a variety of names. It may also be known as adenoameloblastoma and can be differentiated histologically from simple ameloblastoma. The pathogenesis of the odontogenic adenomatoid tumour has been controversial issue since Stafne (1948) originally suggested that the cell of origin was that of epithelium entrapped in the lines of embryonal fusion.

Stafne considered that one his original cases arose from the lining of a globulomaxillary cyst. This theory is unacceptable since it does not explain how the odontogenic adenomatoid tumour can occur in the mandible. Aisenberg (1953), Cahn (1955) and Oehlers (1956) proposed that the stem cell was from any

epithelium that differentiated into either glandular epithelium, but there is little agreement as to the stage of odontogenesis during which the cell arises.

Spouge (1967) considers the odontogenic adenomatoid tumour to be a benign and orderly proliferation of cells at the enamel organ stage of differentiation, whereas Chambers (1973) presented good data supporting the theory that the stem cell is derived from the epithelial rests of malassez around the apex of deciduous teeth.

Most investigators support this point of view that the lesion is not a tumour but an overgrowth of odontogenic tissue or more properly termed as hamartoma.

Electronmicroscopic studies suggest that this tumour is derived from cells of the inner enamel epithelium at the preameloblastic stage (Smith 1979)

The odontogenic adenomatoid generally appears as an asymptomatic slow growing swelling. Courtney and Kerr (1975) in their large series reported only two patients with pain. The mean age of occurrence has been reported as 18, between the ages of 10 and 21 years. The youngest reported patient was 53 years of age. Two thirds of reported cases have occurred in the females and there appears to be a female predominance up to 30 years of age.

Two-thirds of reported cases have occurred in the maxilla, whereas one third have occurred in the

mandible. A unique feature of this tumour is that between 7.5% and 90% of the reported cases have occurred anterior to the canine tooth. (Courtney and Kerr, 1975, Giansanti So meren & Waldron, 1970). They also reported that 74% of the cases in the literature were associated with impacted or unerupted teeth. Growth is slow and by expansion only. Although most adenoameloblastomas are small in comparison to simple ameloblastomas, large lesion producing displacement of adjacent teeth and involvement of the maxillary sinus and orbit have been described.

Discussion

The adenomatoid odontogenic tumour was recognised by Stafne, who believed that the lesion arose from epithelial cell nests and he coined the term epithelial tumour. Lucas considered this tumour to be of enamel organ origin based on the histology and felt it was a true neoplasm. Since these earlier reports, the clinical and pathological features of this tumour have been well characterised. The adenomatoid odontogenic tumour is now known to be a benign neoplasm which occurs most commonly in patients in the second decade of life. Recurrence is rare. It affects females with greater frequency than males and most often located in the anterior maxilla.

Frequently it is associated with unerupted or impacted tooth. This relationship often results in a preoperative diagnosis of a dentigerous cyst.

It was pointed out by some authors that dentigerous cyst generally surround only the crowns of unerupted teeth, often in a symmetrical fashion when small. Odontogenic adenomatoid tumour frequently extends laterally from one surface of an unerupted tooth and involves a considerable portion of the root. On rare occasions, root resorption is noted.

The vast majority of lesions have been treated conservatively because in most cases they looked like cysts and were presumed to do so.

When compared to the simple ameloblastoma, the odontogenic adenomatoid tumour is striking in clinical

Fig. 3. Microphotograph showing epithelial cells forming duct like structures and polyhedral stromal cells (HE 400 x)

feature and microscopic configuration. Radiographically AQT is observed as unilocular lesions that is frequently associated with an unerupted tooth. The radiograph may reveal peripheral condensing osteitis and the presence of small radiopacities in the central radiolucency. The microscopic picture is characterised by a well encapsulated cystic structure with intraluminal proliferation of epithelium. Conservative surgical excision is curative. Simple ameloblastoma is most frequently noted in the posterior mandible of the patients in the fourth decade. Radiographically the simple ameloblastoma tends to present as multilocular lesion rather than the dentigerous cyst like radiolucency associated with the odontogenic adenomatoid tumour. Microscopically the ameloblastoma consists of proliferating strands and islands of adjacent marrow like spaces and minimal encapsulation.

Attempts at surgical excision, therefore are frequently followed by recurrence, necessitating more radical procedure.

In the present case the age of the patient concided very well with typically reported for an AOT. Its microscopical picture confirmed the presence of AOT. Our patient was male as compared to Giansanti's higher incidence in females. Although the tumour was located in the canine area it was not associated with an impacted tooth. These type of lesion may be explained on the basis of an odontogenic origin. There are few conditions which occur in such a narrow age range in such a restrictive site. many theories on the genesis of the odontogenic adenomatoid tumour have been proposed. This would implicate all aspects of the dental epithelium from the dental lamina at one end of the spectrum to the reduced enamel epithelium at the other end. The only universally accepted concept with regard to origin rests with some aspects of the odontogenic epithelium.

The distinctive clinical radiological and microscopic features with a seemingly absence of recurrences indicate that this is benign lesion with much less potential than the ameloblastoma.

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Congratulation

Dr. Bachi T. Hatiram of Dept of ENT & Head and Neck Surgery TN medical College & BYL Nair Charitable Hospital, Mumbai has received the British Academic Conference Organisation (BACO) Fellowship to attend the 10th British Academic Conference in otolaryngology to be held at Cambridge, UK from 4th to 9th July 1999.