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Adenomatoid Odontogenic Tumor-A Rare Case Report

Dr. C.B. Sharma, Dr. (Ms) Parveen Sharma, Dr. Sukhvinder Saluja

Dr. C.B. Sharma
Professor and Head
Department of Oral & Maxillofacial Surgery
D.A.V. (C) Dental College and Hospital
Yamuna Nagar - 135 001, Haryana, India.

Dr. (Ms) Parveen Sharma
Professor
Department of Oral & Maxillofacial Surgery
D.A.V. (C) Dental College and Hospital
Yamuna Nagar - 135 00, Haryana, India.

Dr. Sukhvinder Saluja
Post Graduate Student
Department of Oral & Maxillofacial Surgery
D.A.V. (C) Dental College and Hospital
Yamuna Nagar - 135 001, Haryana, India.

Corresponding Address

Dr. Sukhvinder Saluja
1092, B-11, Manav Chowk
Hisar Road, Ambala City - 134 001
Haryana, India.

Abstract : *The adenomatoid odontogenic tumour is a unique tumour that comprises only 0.1 per cent of tumors and cysts of the jaw and 3 per cent of all odontogenic tumors (Khan MY et al; 1977)[4]. This is generally considered to affect mainly patients in second decade of life. It occurs with greater frequency in females and is found most often in maxilla and in the anterior regions of the jaws. A benign lesion that does not recur, even after simple enucleation (Giansanti JS et al; 1970)[2]. A case report of a 42-year-old female patient is presented with swelling in the lower anterior region.*

Introduction

Adenomatoid Odontogenic Tumor (AOT) is a distinct odontogenic neoplasm that was first described by Stafne in 1948[6]. This is a well-circumscribed lesion derived from odontogenic epithelium that usually occurs around the crowns of unerupted anterior teeth. This tumor usually affects the younger age group with female: male ratio of 2:1 (Lee JK, Lee KB, Hwang BN; 2000)[5]. Radiologically, it is characterized by a unilocular radiolucency, which is generally associated with unerrupted tooth (Courtney RM, Kerr DA; 1975)[1]. Histologically the tumor consists of encapsulation around odontogenic epithelium, which is arranged in ductal and swirls patterns interspersed with spherical calcifications.

Case Report

A 42-year female patient reported to Oral and Maxillofacial Department of D.A. V (C) Dental College and Hospital with a chief complaint of swelling in the front region of the lower jaw of 2 months duration. The swelling started 5 years back, and gradually increased to the present size and had a completely asymptomatic course without any interference with mastication, speech and esthetics. She lost her four anterior teeth during the course of swelling.

Medical history revealed the patient to be a controlled hypertensive. She was otherwise fit and with all vitals within normal limits. Personal and family history was unremarkable.

Extra oral examination showed a swelling in mandibular anterior region without any sinus/fistula. The overlying skin was of normal color and texture and not fixed to the underlying structures (Fig. 1).

The swelling was firm and non-tender on palpation. There was no associated paresthesia of lip and chin region. Mouth opening and TMJ movements were within normal limits.

Intraorally, a non-tender, firm swelling was seen extending from right mandibular 1st premolar to left mandibular 1st premolar region of approximately 4cm x 3cm with distinct margins. The overlying mucosa was of normal color and texture with no evidence of ulceration (Fig. 2).

There was no sign of bruit or pulsation. There was gross buccal and lingual cortical plate expansion. Patient had lost her mandibular central and lateral incisors of both sides during the course of swelling and her right mandibular canine was grade III mobile.

Patient's routine blood and urine investigations were within the normal limits. She was also investigated for serum calcium, phosphate, alkaline phosphatase levels and renal function tests, which were also found to be within normal limits.

Radiographic examination included an OPG (Fig. 3) and lower occlusal film (Fig. 4), which showed, a well-circumscribed mixed radiolucent radiopaque lesion in anterior region of mandible measuring 3 x 4cm in its greatest extent with marked buccal expansion. The right mandibular canine was present at the top of the lesion, which was running 1cm short of the inferior border of the mandible.

Provisional Diagnosis: On the basis of clinical history and radiographic examination, a provisional diagnosis of ossifying fibroma was considered.

Fig. 1 : Extra oral Photograph lateral view

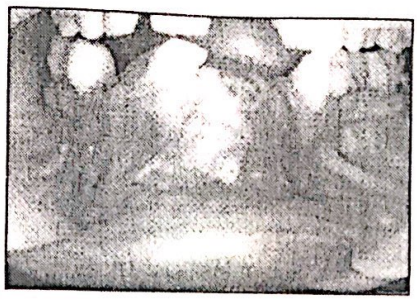

Fig. 2 : Intra oral view

Fig. 3 : Preoperative orthopantamogram

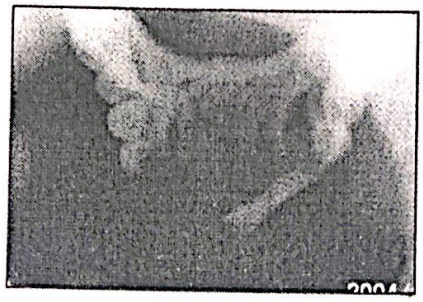

Fig. 4 : True occlusal view

Differential Diagnosis:

The differential diagnoses included were as: ossifying fibroma, desmoplastic ameloblastoma and adenomatoid odontogenic tumor.

Ossifying fibroma was considered as most often occurs over a wide age range with the greatest number of cases encountered during the third and fourth decades of life with a definite female predilection. Mandible is involved far more than the maxilla but most often noticed in the premolar molar region of mandible in comparison to anterior region.

This tumor is usually asymptomatic, but later on produces expansion of the jawbones. On X-ray, a well-demarcated radiolucent area is seen in the early stages, which passes through the different stages of mixed radiolucent/radiopaque to the final radiopaque stage (Neville BW, Damm DD, Allen CM, Bouquot JE; 2002)[9].

Desmoplastic ameloblastoma was included as it is most likely to occur in the anterior or premolar region of the jaws and approximately half of the desmoplastic lesions are located in the maxilla, there is no difference in prevalence between the maxilla and mandible.

Cases have been reported in patients aged 18 to 70 years with a mean of 41.2 years. No difference between sexes has been reported. On contrary, the incidence of desmoplastic ameloblastoma is low; rates of 0.9% to 12.1% of all ameloblastomas.

The radiographic appearance of this neoplasm usually indicates a mixed radiolucent/radiopaque lesion. Approximately half of these lesions have diffuse borders in the radiograph and look similar to a fibro-osseous lesion or malignant tumor (Kaffe I, Buchner A, Taicher S; 1993)[3]. The lamina dura also is involved (Tanimoto K, Takata T, Suei Y, Wada T; 1991)[10].

The adenomatoid odontogenic tumour is slow growing painless tumor, which characteristically affects the anterior region of the jaws in usually second decade of life with more female predilection than male (Giansanti JS et al; 1970)[1]. The maxilla is involved nearly twice as frequently as the mandible. Radigraphically, it is usually characterized by a unilocular radiolucency associated with unerupted teeth and sometimes multilocular also.

The case reported was of considerable interest to us because of similarity to other lesions, which give similar clinical and radiographic presentation. The most likely lesions were ossifying fibroma and desmoplastic ameloblastoma. Age, sex and radiographic features are very similar to ossifying fibroma but site was not favorable in this case. Age and site of the present lesion were similar to desmoplastic ameloblastoma but the incidence of desmoplastic ameloblastoma is low, 0.9% of all ameloblastomas and radiographically desmoplastic ameloblastoma has diffuse border in contrast to well defined border in the present lesion. Other lesions like giant cell granuloma and glandular odontogenic cyst were also considered but were ruled out for lack of favourable findings.

Surgical Procedure

Surgical excision of the lesion was planned because of non-aggressive nature of the lesion, under general anesthesia. The lesion was approached through an intra oral approach and a full thickness mucoperiosteal flap was raised from 35 to 45 regions (Fig. 5).

Extraction of mobile left mandibular canine was done, The tumor mass was shelled out easily from the underlying bone and surrounding mucosa. After excising the mass, hemostasis was achieved and iodoform pack was given and flap was sutured with 3-O vicryl sutures (Fig. 6 and 7).

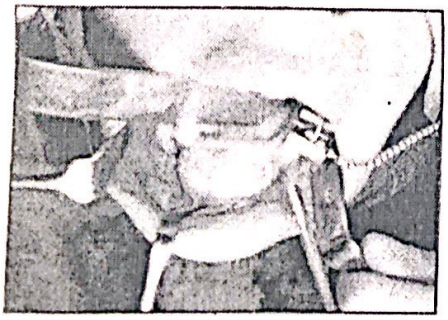

Fig 5 : Intra operative photograph showing raised mucoperiosteal flap

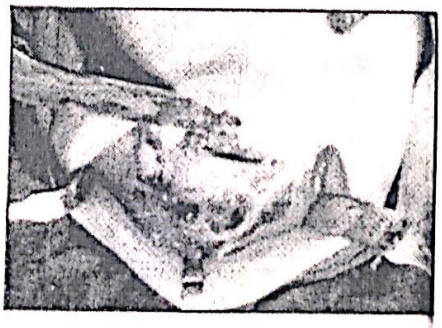

Fig. 6 : Intra operative photograph showing tumor mass excised

Fig. 7 : Intra operative photograph showing closure done.

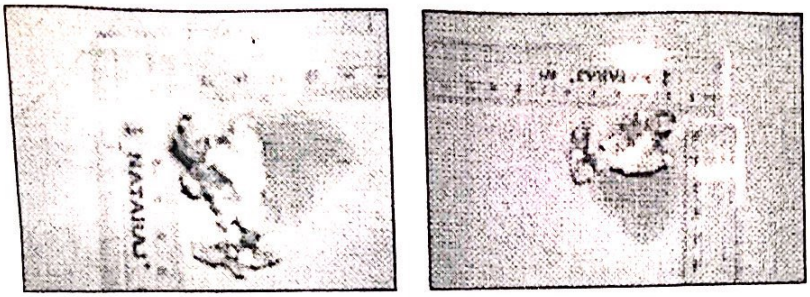

Fig 8 A B : Excised lesion

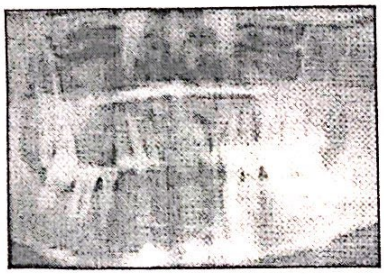

Fig. 10 : Post operative orthopantamogram 6 months

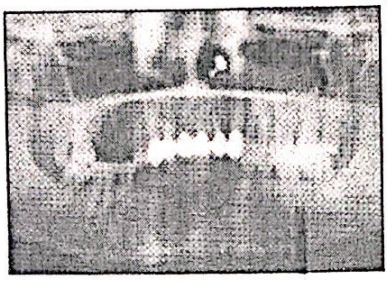

Fig 11 : Post operative orthopantamogram 1 year

Specimen, sent for histopathological examination was found to be an adenomatoid odontogenic tumor (Fig. 8A and 8B).

Histopathological Report

Section stained with eosin and haematoxylin stain revealed a lesion surrounded fibrous connective tissue capsule. The lesion revealed the presence of odontogenic epithelial cells, which were arranged in sheet and islands. In sheets, the odontogenic epithelial cells formed duct like areas, which were surrounded by spindle shaped epithelial cells. Different foci of calcifications were seen throughout the stroma. The overall histological picture was suggestive of adenomatoid odontogenic tumor (Fig 9A 9D).

Subsequent Course

Postoperative orthopantomogram after 6 month (Fig. 10) and 1 year (Fig. 11)

showed satisfactory bone formation. Patient is under regular follow up and doing well. (Fig. 12 A D).

Discussion

Adenomatoid odontogenic tumor was initially thought to be a variant of ameloblastoma and was therefore referred to as ameloblastic adenomatoid tumor or adenoameloblastoma. In 1969, Phillipsen and Birn[3] suggested the term AOT, which is accepted today.

AOT possess characteristic clinical features that render the diagnosis relatively obvious. First this tumor is largely limited to young patients, with two thirds of all cases occurring between 10-19 years of age. Second, its occurrence is almost always confined to the anterior region of the jaws. Third, there is a decisive sex predilection for females, with the ratio of about 2:1(Lee JK et al; 2000)[5]. Our case reflects the second and third features but not first. In our case the age of patient is 42 years old.

Lesions are typically asymptomatic, and adjacent teeth may be displaced. Large lesions can cause painless hard swellings, as in this case.

Radiographically, the AOT comprise a follicular and extrafollicular type. The follicular type shows a well defined, unilocular (round or oval) radiolucency associated with crown and often part of the root of an unerrupted tooth thus mimicking a dentigerous cyst (Phillipsen HP, Reichart PA; 1998)[6].

The extrafollicular type is, on the other hand not associated with an unerrupted tooth and well defined unilocular radiolucency is found between, above or superimposed upon the roots of erupted permanent teeth. (Phillipsen HP, Reichart PA; 1998)[4]. In our case, the AOT is extrafollicular variety. In approximately 2/3rd of the intrabony variants the radiolucency shows discrete foci having floceulent pattern of scattered radiopacities. In the present case, the lesion showed mixed radiolucent/ radiopaque image owing to calcifications present.

Histologically , the tumor is composed of spindle shaped epithelial cells that form sheets, strands, or whorled masses of cells in a scant fibrous stroma. The epithelial cells may form rosette like structures and tubular or duct like pattern may be prominent or scanty. Small foci of calcification may also be scattered throughout the tumor. Duct like spaces are lined by a single row of low columnar epithelial cells, the nuclei of which are polarized away from the lumenal surface (Phillipsen HP, Reichart PA; 1998)[6].

Since AOT shows benign clinical behavior and is well encapsulated, conservative surgical enucleation or

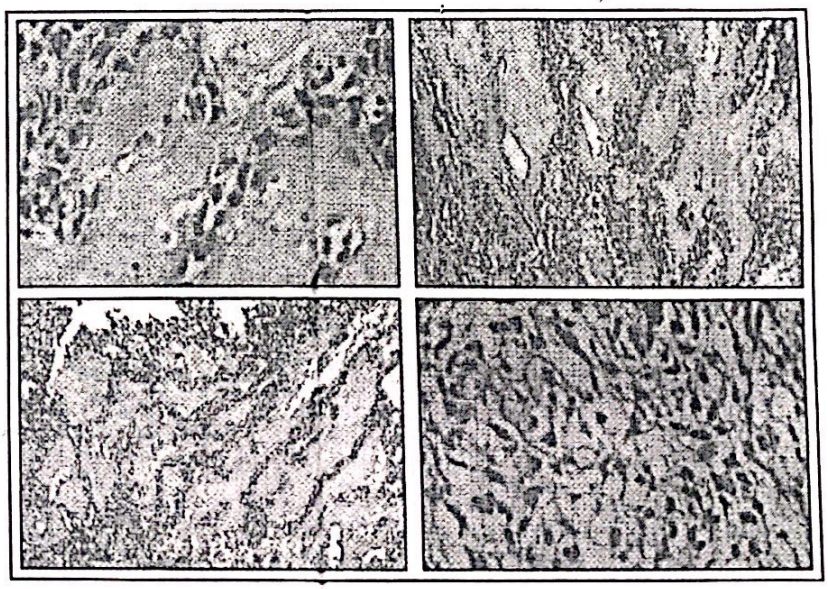

Fig 9 A-D : Histopathological Sections

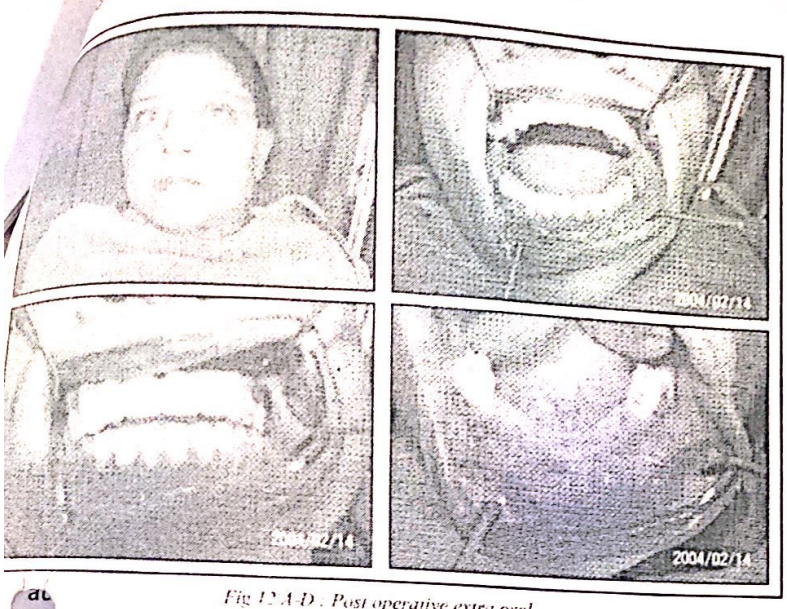

Fig 1 2 A-D : Post operative extra oral and intra oral photograph

ettage has proven the treatment dality of choice. Recurrence rate is y low (Phillipsen HP, Reichart PA; 8).

nclusion

e a rare case report of 42 year female ent with adenomatoid odontogenic or of anterior mandible has been ented, which otherwise occurs more monly in younger patients and in illa. Recovery of patient was ventful and patient is still under w up.

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