Ameloblastic fibroma in the midline of mandible: a case report

P. K. Mohapatra* / A. R. Choudhury** / H. Parkash***

The ameloblastic fibroma (AF) is a rare benign mixed odontogenic tumor. It is composed of both epithelial and mesenchymal elements, but lacks of any calcified dental structures. Most of these tumors occur in the mandible and appears preferentially in the posterior portion of the dental arch with molar area predominates over premolar area. It is important to differentiate the lesion from ameloblastoma, since unlike the latter, it does not exhibit a locally invasive growth pattern. It is a well-circumscribed lesion and does not require the radical excision that may be necessary to effect cure with ameloblastoma. The present case report describes a 15-year-old patient with an ameloblastic fibroma in the symphysis of the mandible, a rare reported site. In the beginning of the article an extensive review of the previously published literature on ameloblastic fibroma has been made. In the later part, the diagnosis, differential diagnosis, histology and therapeutic procedures and postoperative follow up of the present case have been described.

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INTRODUCTION

Odontogenic jaw tumors in which varying degrees of both dentin and enamel matrix (with or without calcification) occur are of two types. The "ameloblastic fibroma" is a tumor of children, which resembles ameloblastoma radiographically and histologically (except that the stroma consists of pulp tissues rather than undifferentiated connective tissues) and which behaves less aggressively than ameloblastoma. The other type comprises various odontomas such as ameloblastic fibro-odontoma and complex and compound odontomas.[1] Ameloblastic fibroma (AF), a true mixed tumor, is a relatively uncommon neoplasm of odontogenic origin which is characterized by the simultaneous proliferation of both epithelial and mesenchymal tissues without formation of enamel and dentin.[1-4]

* Dr. P. K. Mohapatra, Senior Resident and Ph.D. Student, Department of Dental Surgery, All India Institute of Medical Sciences, Ansari Nagar, New Delhi 110 029, India.

** Dr. A. R. Choudhury, Assistant Professor, Department of Dental Surgery, All India Institute of Medical Sciences, Ansari Nagar, New Delhi 110 029, India.

*** Dr. H. Parkash, Professor and Head, Department of Dental Surgery, All India Institute of Medical Sciences, Ansari Nagar, New Delhi 110 029, India.

Address all correspondence to Dr. Han Parkash, Professor and Head, Department of Dental Surgery, All India Institute of Medical Sciences, Ansari Nagar, New Delhi 110 029, India.

Phone 6593231, 6864851 ext. 3231 (0) 6594549 (R)
Fax: 6862663
E-mail hariparkash@hotmaiI.com

It was first described by Kruse[5] in 1891 and reported to constitute 2.5% of total odontogenic tumors.[5-7] The AF occurs predominantly in a younger age group than the usual ameloblastoma.[6] However, it has been reported that this tumor can occur at an age ranging from 6 months to 42 years[8] with 70% of it occurs under 20 years of age.[6,9] (with an average age of 14.61 to 15.5 years[10]). The sex predilection reported to vary from no preference,[6] to a male female ratio that can vary from 2:1[11] to 4 : 4.3[12] Although this tumor may arise in either jaw, the majority occurs in the mandible,[13,14] but percentages vary from 80%[5] and 83% to 90%.[6] The posterior portion of the dental arch is the location for virtually all ameloblastic fibromas, with the molar area favored over the premolar area in its location.[4,6] After an extensive review of published literatures we could find reports of only five anterior maxillary ameloblastic fibroma.[8,10,12-15] However, we are not able to find reports of any case of this tumor in symphysial area of mandible.

CLINICAL FEATURES

These lesions are usually asymptotic and are discovered incidentally (17%), or because of growth causing expansion (58%)[4,5,10,13] The tumor can vary in size from 1 to 8.5 cm.[4,6,10] They grow slowly by expansion of the cortex and are reported to enlarge by extension as a solid mass.[4,15] This tumor usually does not invade bone[4,6-14] and not aggressive.[10] It may produce enlargement, expansion of the cortex of the jaws, and in some case displacement of teeth.[14]

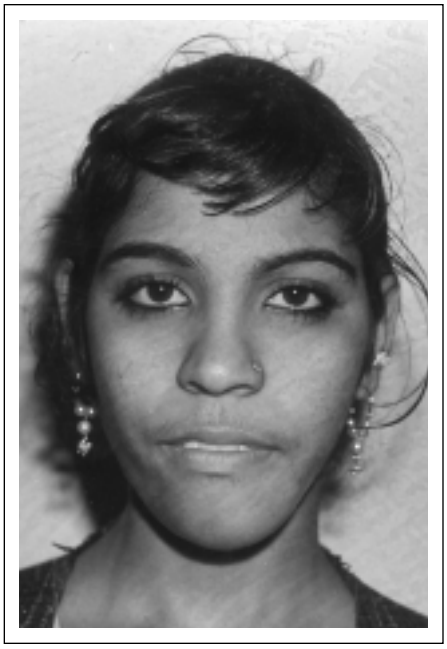


Figure 1. Pre treatment extra oral view showing marked prominence of chin secondary to underlying swelling.

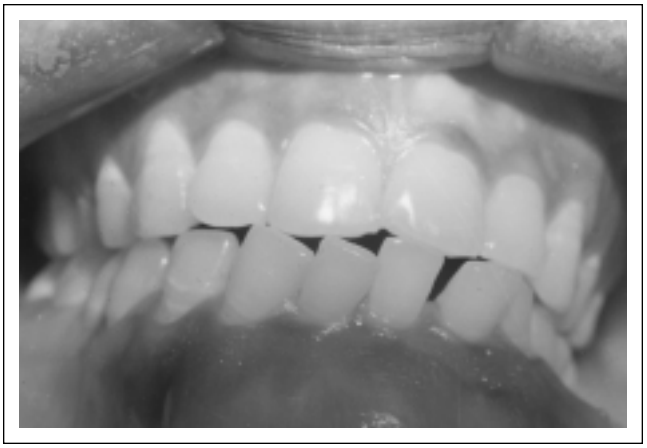


Figure 2. Pre treatment intra oral view showing the extent of the lesion with pathological migration of lower anteriors.

RADIOGRAPHIC FEATURES

The roentgenographic features are characterized by a well-defined radiolucency, which are either unilocular or multilocular.[4,10,13,15,16] It may be associated with an unerupted tooth[12,17] when it is usually seen as pericoronal radiolucency.[14] It may show cortical expansion and spread the roots of adjacent teeth and/or their displacement.[10,14,18]

HISTOLOGICAL FEATURES

Histologically, the AF is often encapsulated and contains both ectodermal and messenchymal elements.[8] Mitotic activity is sparse, and there is no calcified tissues.[4-9] The tumor usually consists of proliferating strands and clumps of odontogenic epithelium lying in highly cellular fibroblastic tissue stroma resembling dental papilla of the developing tooth.[19] The odontogenic ectodermal (epithelial) component shows a peripheral layer of cuboidal or columnar cells in strands, cords or nests. The nests may have central areas that resemble stellate reticulum. The appearances are similar to ameloblastoma, but the stellate cells are much less abundant and the cyst formation is unusual.[4,6,8]

The cell rich mesenchymal component consists of a loose, primitive mesenchymal tissue, often contains stellate nuclei[4,6,8] The messenchymal component is markedly differentiated from the fibrous stroma of ameloblastoma. It resembles the tissue forming dental papilla of the tooth germ. There may be a narrow cell-free zone of hyaline connective tissue border around the epithelial component.[19]

In some lesions, dentine and enamel matrix may be present and such tumors may be diagnosed as ameloblastic fibro-odontoma or fibrodentinoma (in absence of enamel matrix. Whether or not they are distinct from the AF are not clear.[19] Because of the cellularity of the messenchymal component of the AF, it gives the impression of a formidable neoplasm. It does not malignant. The tumor infiltrates the marrow spaces as a solid mass and therefore is not so insidious as the ordinary ameloblastoma, which infiltrates in cords, strands and finger like process.[16]

TREATMENT

Since the ameloblastic fibroma is often encapsulated and are reported to enlarge by extension as solid mass without infiltrating the marroe spaces; hence the treatmet for the initial lesion is generally recommended to be conservative, i.e., simple excision,[16] enucleation[14] or curettage.[6] The tumor readily separates the bone and ameloblastic cells generally do not invade the connective tissues capsule. Long term (at least 10 years) follow up evaluations indicating no recurrence.[4,7,12,15] However, Zallen *et al*[2] have reported that the recurrence rates are higher (approximately 18%) for this tumor than for the adenomatoid odontogenic tumor (previously known as adenoameloblastoma).[2]

CASE REPORT

A 15 year old girl reported to the Out Patient Department of Dental Surgery, All India Institute of Medical Sciences, New Delhi with chief complaints of swelling in the frontal region of the lower jaw since 3 years and drifted and loose of lower front teeth for one year. The present history of the patient revealed that the swelling was slowly growing over the time and was not associated with pain or discharge. However, she had distorted lower third of the face due to increased size of the swelling (Figure 1).

The past history and medical history was unremarkable. She was taking no medication and had no history

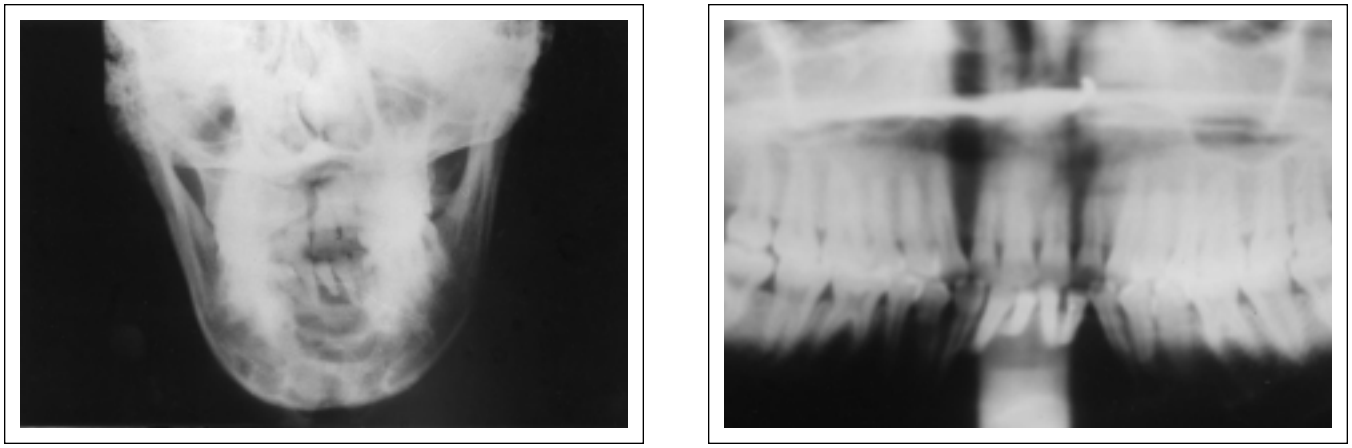


Figure 3. Pre treatment radiographs (A) PA view of mandible, (B) Panoramic view showing large well-circumscribed radiolucent lesion in the symphyseal area of the mandible. There is marked root resorption of lower incisors.

of known drug allergy. Her physical examination showed no abnormalities other than those related to the chief complaint. On extra-oral examination, she had enlarged symphysis with a hard swelling of approximately 5 cm x 5 cm size extending approximately from mental foramen of one side to the same anatomical location of the other side anteroposteriorly and from labio-mental sulcus to the lower border of the mandible supero-inferiorly. The swelling was non-tender, non-compressible, non-reducible, non-fluctuant and the temperature on the swelling area was normal. There was no extra-oral sinus and there was eggshell crackling in the central area of the swelling. The skin overlying the swelling was free and mobile.

Intra-orally, most of the swelling was on the labial aspect of the arch obliterating the labial sulcus. There was a gross expansion of the symphysis with a large swelling of approximately 6 cm x 5 cm size extending from first premolar of one side to the first premolar of opposite side of the arch antero-posteriorly and from free gingival margin superiorly to an ill defined extent inferiorly. The swelling was causing a downward displacement of lower lip. The patient had a full complement of teeth extending from permanent central incisors to third molars in all the quadrants of both the arches without having any carious or filled component. However, there was drifting and grade II mobility of mandibular central and lateral incisors of both the sides towards midline in the swelling area. The periodontal status was fair with a normal probing depth in the swelling area. The mucosa overlying the swelling was thin and mobile with a bluish hue of the overlying tissues (Figure 2).

On palpation the swelling was large, firm, expansile mass and extending on both the sides of the mandible crossing the midline. There was massive expansion of labial cortical plate and a minimal expansion of lingual cortex. The eggshell crackling was positive in certain areas of swelling without signs of any fluctuation. There was no pulsation, bruits or abnormal palpable warmth over the swelling area. The remainder of the examination was negative.

All the laboratory investigations including blood chemistry were within normal limit.

RADIOLOGY

The postero-anterior radiograph mandible (Figure 3A) and the panoramic radiograph (Figure 3B) revealed a large unilocular radiolucent lesion with a well defined margin extending from permanent canine of one side to the permanent canine of the opposite side. There was extensive resorption of roots of all the lower incisors with adequate basal bone in the lower border of symphysis.

After analysing the clinical findings and radiological features of the lesion, the case was diagnosed provisionally as a case of ameloblastoma with a differential diagnosis of ossifying fibroma, fibrous dysplasis, trabecular adenoma, ameloblastic fibroma, ameloblastic fibrosarcoma and intraosseous adenoid cystic carcinoma. To confirm the diagnosis before going for definitive surgical approach a fine needle aspiration biopsy (FNAB) of the lesion was performed.

FINE NEEDLE ASPIRATION BIOPSY (FNAB)

Aspiration of the tumor yielded a cellular sample composed of a mixture of mesenchymal and epithelial cells. The mesenchymal component consisted of loosely arranged fusiform cells, arranged in thick mass where as the epithelial component was represented as complex solid structures (clusters) whose borders were columnar cells with pallisading and the central regions reminiscent of stellate reticulum. These cytological features appear to be sufficiently characteristic to suggest a diagnosis of ameloblastic fibroma.

After the FNAB report was made available the patient was scheduled for enucleation of the lesion fol-

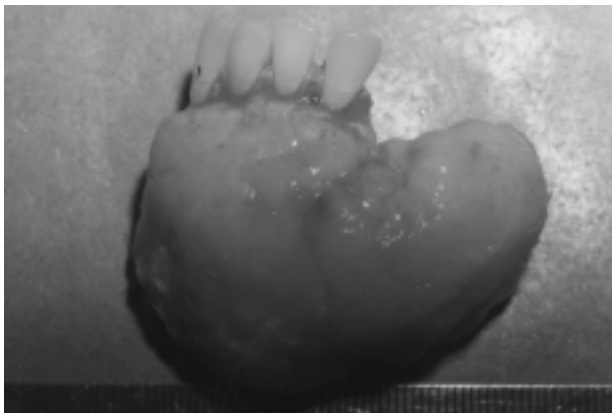


Figure 4. The excised tumor in toto.

lowed by histopathological examination of the removed tissues for a confirmed diagnosis.

SURGICAL PROCEDURES

General anesthesia was induced with naso-tracheal intubation. After the induction of anesthesia the skin as mucosa was prepared in a conventional way. Xylocaine 1% with 1:80,000 adrenaline was used for hemostasis and local anesthesia. An envelope type incision with lateral releasing was made. The incision was carried through mucosa and careful dissection around the lesion was established with the help of a free elevator along a plane between the lesion and overlying mucosa. A mucoperiosteal dissection was accomplished to free lateral margins of the tumor. Then the overlying thinned out labial cortical bone was removed. The dissection was carried inferiorly to expose the most part of the tumor. The lesion was removed intact, after separating its lingual attachment along with all four central incisors. Since there was an extension of the lesion from permanent canines on both sides, both the canines were extracted. The removed lesion appeared to be firm and well capsulated. It was believed that there was no need for frozen sections because the lesion appeared to be removed entirely. However, the remaining part of the bony wall of the lesion was thoroughly curretted with an aim to remove any extended part of the tumor down to the bone. Finally the area was irrigated and debrided with gentamycin wash followed by Betadine. The incisions were closed primarily. There was no significant bone loss and the recovery of the patient from general anesthesia was uneventful.

SURGICAL PATHOLOGY

The tumor measured 5.5 x 5.0 x 3.5 cm in its largest dimension with the cut surface showed a round, well circumscribed, grayish myxoid mass surrounded by a thin transparent capsule (Figure 4).

The microscopic examination revealed neoplastic tissues characterized by islands of epithelial cells arranged in long finger like strands, cords and nests. The epithelial cells were cuboidal to columnar and well differentiated. The islands of epithelium were separated by an immature myxoid stroma. Mitotic figures were not seen. The border was not infiltrative and the neoplasm was focally rimmed by a well vascularized, loose connective tissue. A final diagnosis of ameloblastic fibroma was made (Figures 5, A-C).

POST OPERATIVE COURSE

The patient did well post-operatively and was sent home after seven days of hospitalization. She was instructed to maintain proper oral hygiene. The surgical site healed without any break down (Figures 6, A & B). After three weeks the restorative care was done by prosthodontist by providing a clasp retained removable partial denture (Figure 7, A & B). She was kept under follow up observation for any recurrence in 3 to 4 months interval and three years after surgery there was no clinical signs of recurrence. She will continue to be reviewed annually.

DISCUSSION

The discussion regarding any swelling along the midline of the mandible centers around three important issues, i.e. proper diagnosis (type of tissue origin), mode of treatment and the potential of the lesion to recur either as malignant lesion or benign one. In regard to the first question, after reviewing the detail history, clinical and radiological findings it is possible to diagnose the case as a chronic lesion or an acute one. In case of ameloblastic fibroma the lesion is chronic, slow growing with healthy teeth in the involved area. However, in case a large growth the teeth in the involved area may be drifted and mobile as in the present case report. The radiographic examination is also helpful to rule out any periapical periodontal pathology and also the characteristic of an odontogenic tumor. However, before proceeding for a definitive treatment it is always advisable to have a fine needle aspiration biopsy (FNAB) of the lesion in order to establish a proper diagnosis, which is not only useful for the clinician but also for the patient as the line of treatment (approach to eradicate the lesion) varies from lesion to lesion. So far as the ameloblastic fibroma is concerned the FNAB findings is characteristic, as described earlier.

There are certain conflicting data in the literature regarding recurrence rates, as well as whether the recurrent tumor is actually a recurrence or residual tumor left at the time of the initial surgery. In medicine the definition of residual is "remaining or left behind" where as recurrent means "returning after intervention or remission."[20] When these definitions are combined, it would seem logical that all recurrent lesions are a result of residual tumor. The idea of a second primary lesion

in the same area seems to be very unlikely. Many authors state that tumor removal was clinically accomplished, but it usually can not be stated that all tumor was removed at a cellular level. Hence, we conclude that all the recurrent lesions are from residual tumors.

The behavior of odontogenic tumors, in particular the ameloblastic fibroma, with time remains a subject of controversy. Zallen *et al.*[2] after an extensive review of literature on ameloblastic fibroma concluded that 14 cases out of 82 cases reviewed had recurrences for a rate of 18.3%. Gorlin *et al.*[11] reviewed the clinical behavior of 35 cases in which there was recurrence in 2 cases (5.7%). Trodahl[10] surveyed 24 cases out of which he found recurrences in 10 cases (43.5%) and Regezi *et al.*[21] reviewed 15 cases in which they did not find any recurrence (0%). Therefore, we are of the opinion that probably the initial ameloblastic fibromas had not been removed completely or it is also likely that recurrent lesions might have occurred but have not been reported.

In regard to the malignant potential of the ameloblastic fibroma, Muller *et al.*[22] reviewed the literatures on the ameloblastic fibromas comprised of 51 cases. These are the cases which was shown to develop *de novo* or in recurrent ameloblastic fibromas or in ameloblastic fibro-odontomas. They concluded that the ameloblastic fibrosarcoma is histologically a malignant tumor, but its biologic behavior is unlike a malignant, that is, only 1 of the 51 cases is reported to have metastasized. After comparing the nuclear content of 5 ameloblastic fibrosarcomas and 3 ameloblastic fibromas by image analysis, they concluded that there was no correlation with histological grade and DNA aneuploidy. This is in contrast to the findings in many histologic types of sarcomas where there is a passive relationship between increase in histological grade and DNA aneuploidy. In addition to it, Chomette *et al.*[23] by histo-enzymological study found a high level of alkaline phosphatase and ATPases activities in all of the samples, a feature not present in common fibrosarcomas. They also found that the histological documentation of 44% of ameloblastic fibrosarcomas (19 out of 43) cases were reported to be developed from pre-existing ameloblastic fibroma.

So far as the management of initial lesion of ameloblastic fibromas is concerned, there are two schools of thoughts. One school, in view of the histological documentation that 44% of the ameloblastic fibrosarcomas developed from pre-existing ameloblastic fibromas and a cumulative recurrence rate of 18.3%, advocates an initial block resection when there is a diagnosis of ameloblastic fibroma in frozen section or return of such a histological diagnosis on the regular sections lesion.[22] According to the proponents of this concept enucleation and/or curettage seems likely to lead to incomplete removal of the tumor tissues. However, Zallen *et al.*[2] also state that they would not always

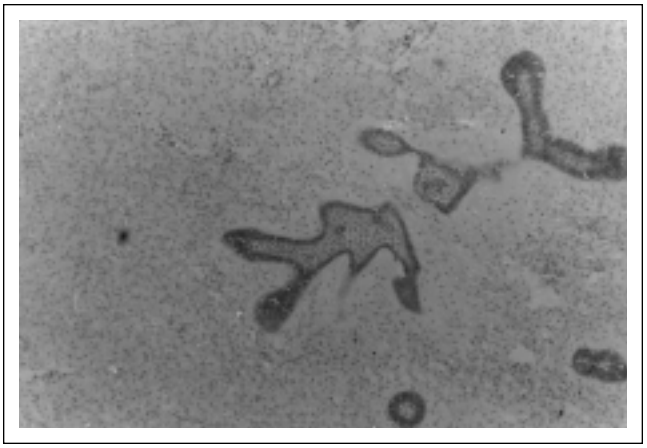


Figure 5. Photo micro-graph of the micro-section of excised tumor after Haematoxylline and Eosin stain. A. Showing a cell rich mesenchymal stroma containing both small islands and long branching cords of odontogenic epithelium (Original magnification X 25).

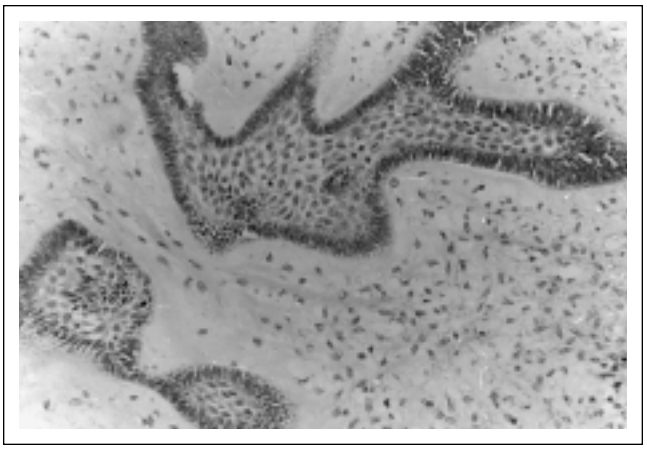


Figure 5. Photo micro-graph of the micro-section of excised tumor after Haematoxylline and Eosin stain. B. Showing branching epithelial cords with peripheral columnar cells surrounding a central stellate reticulum like tissues. The cellular stroma resembles the dental papilla (Original magnification X 100).

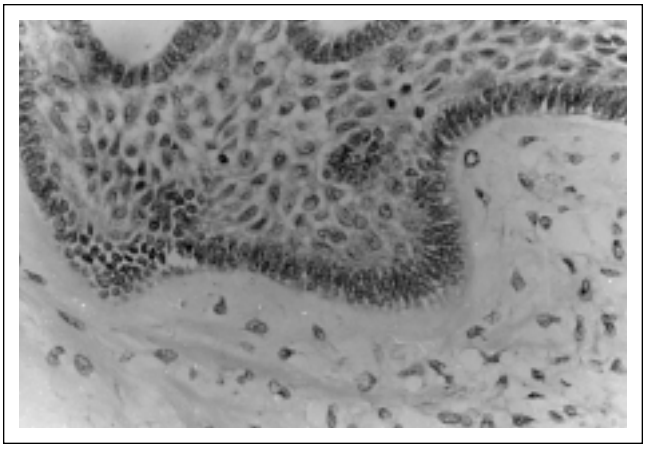
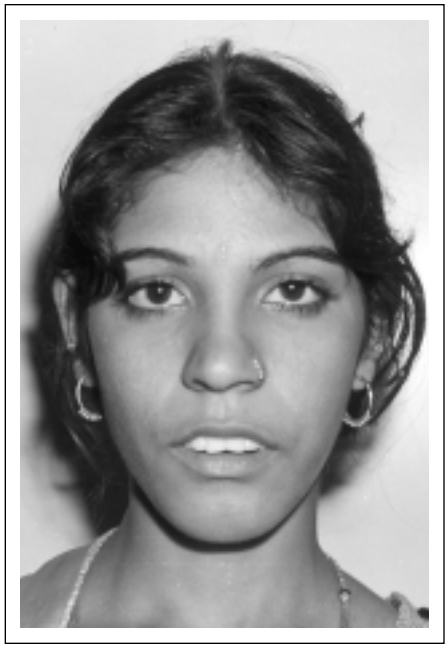


Figure 5. Photo micro-graph of the micro-section of excised tumor after Haematoxylline and Eosin stain. C. Showing epithelial islands lined by columnar cells exhibiting reverse polarity. The mesenchymal stromal cells are ovoid and some times stellate. Note the lack of collagen that is typically present in ameloblastoma (Original magnification X 400).



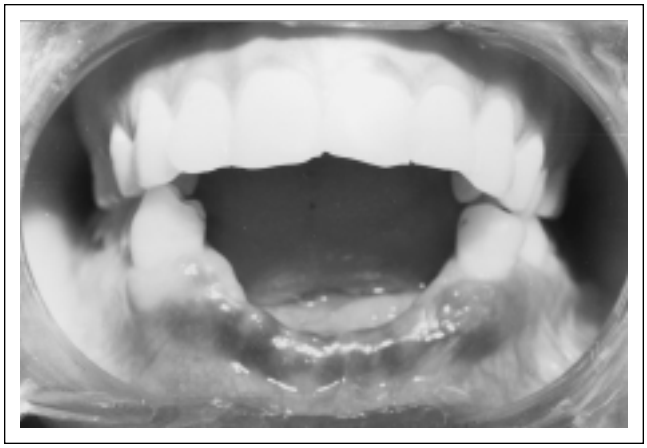


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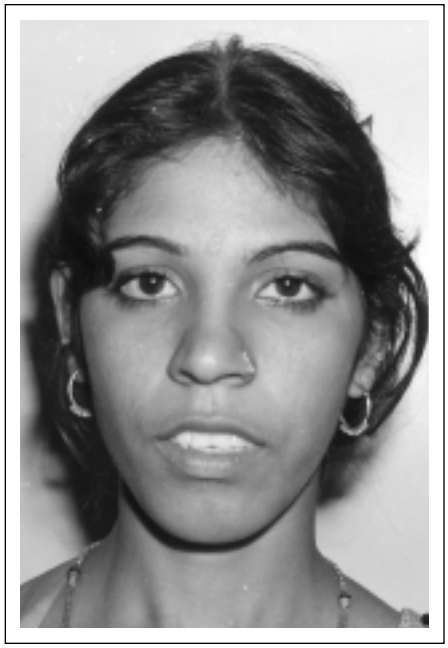
Figure 6. Post surgical view after three weeks (A) extra oral view, (B) Intra oral view showing properly healed surgical site.



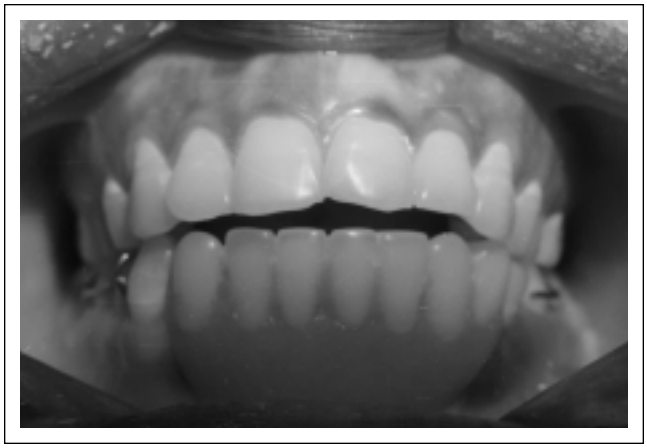


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Figure 7. Clinical view after prosthetic rehabilitation (A) extra oral view, (B) intraoral view.

follow the recommendations of a modified block resection if it would result in sever disfigurement. The other school of thought believe in the fact that in case of an initial ameloblastic fibroma lesion conservative approach including enucleation, mechanical curettage with good follow up and then modified resection if there is any recurrence is more logical.[8,24-27]

After evaluating the literature we are of the opinion that a conservative approach should be adopted for the initial lesions of ameloblastic fibroma. However any recurrent lesion should be treated with modified block resection and in all cases a good follow up protocol (at least review once in a year) must be followed.

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