Highly Proliferative Ameloblastic Fibroma: A Rare Entity

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Abstract

Ameloblastic fibroma (AF) is a rare odontogenic neoplasm which is commonly seen in the second decade of life. It is seen most frequently in the mandibular posterior region. AF shows clinical and radiographic resemblance with other commonly occurring odontogenic cyst and tumors. Histopathologically, it shows great resemblance with primitive dental papilla. Immunohistochemistry helps in understanding the nature and proliferative potential of tumor and helps in proper treatment planning. Large lesions and recurrent lesions are treated with segmental resection which can often lead to morbidity, especially in young patients if not managed properly. Herein, we present a case of a large AF in the posterior mandible region in a 21-year-old female patient with significant expansion and erosion of cortical plates and lower border of the mandible with a high Ki67 proliferative index (20%) which was surgically treated by segmental resection and immediate reconstruction by autogenous iliac graft.

Keywords: *Immunohistochemistry, mandibular reconstruction, neoplasm, therapeutic use*

Introduction

The WHO defined ameloblastic fibroma (AF) in 2005 as "Neoplasm consisting of odontogenic ectomesenchyme resembling the dental papilla and epithelial strands and nests resembling dental lamina and enamel organ. No dental hard tissues are present."[1] It was first described in literature in 1891 by Kruse.[2] In 1971, AF was first classified as benign neoplasm, and in 1992, the WHO classified it as mixed neoplasm.[3,4] AF is relatively uncommon neoplasm consisting of 1.5%–4.5% of all the odontogenic tumors occurring frequently in the second decade of life.[3,4] Some believe AF to be a slow-growing innocuous tumor while some suggest it to be more aggressive tumor which may be a part of a spectrum of lesions which mature from AF to ameloblastic fibro-odontoma to odontoma.[5,6] The present case highlights the importance of careful diagnosis of intrabony oral lesion in a young patient and usage of immunohistochemistry (IHC) in understanding the nature of tumor and selecting appropriate treatment option.

Case Report

A 21-year-old female reported to the hospital with a history of asymptomatic progressive facial swelling for 1 month. Extraorally, slight facial asymmetry was noted on the left side [Figure 1a]. Intraoral examination revealed hard, diffuse swelling in the mandibular body from second premolar to second molar region with expansion of buccal and lingual cortical plate covered by normal mucosa [Figure 1b and c]. No paresthesia or pain associated with the swelling. The panoramic radiograph showed the presence of a well-defined multilocular radiolucency involving the left mandible with root resorption of the teeth. A cone-beam computed tomography scan showed an expansive lesion with 8.0 cm × 3.0 cm × 4.5 × cm in extension causing loss and buccal and lingual cortical plates and invasion of lesion in the mandibular canal and lower border of mandible in the molar region [Figure 1d and e].

Following incisional biopsy, the specimen was sent to histological examination. Microscopically, ameloblastomatous islands were noted with central stellate reticulum cells enmeshed in fibrocollagenous stroma which resembled primitive dental papilla. Proliferating odontogenic epithelium cells from the discrete island exactly resembled with the follicular stage of dental papilla with centrally placed stellate reticulum cells. Connective tissue mesenchyme consisted of plump stellate and ovoid cells in loose matrix [Figure 2].

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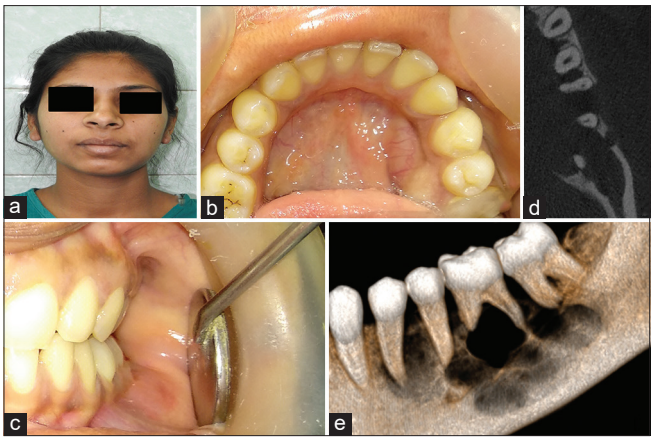

Figure 1: Clinical and cone-beam computed tomography images. (a) Preoperative frontal image of a patient showing mild facial asymmetry on the left side. (b) Intraoral image showing swelling in the lingual region with respect to 44, 45. (c) Intraoral image of the buccal vestibule showing obliteration of the left buccal vestibule in the molar region. (d) Transverse cone-beam computed tomography section of the mandible at mid-root region showing extensive bone expansion and perforation of buccal and lingual cortical plates. (e) Three-dimensional reconstructed image of buccal aspect of the mandible showing invasion of inferior alveolar canal and loss of cortical plates

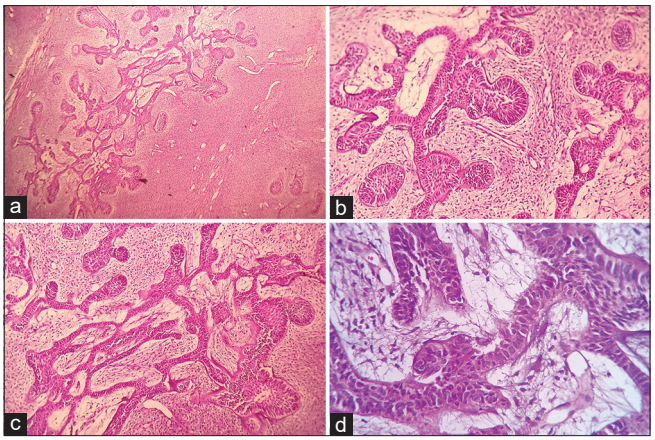

Figure 2: Histopathological examination. (a) Histopathological image showing ameloblastic islands in cellular connective tissue stroma. (b) ×4 view. (c) ×10 view. (d) Tall columnar cells with reversal of polarity resembling primitive odontogenic epithelium

The diagnosis of AF was established. Despite being locally aggressive, considering the high possibility of recurrence, segmental resection of the mandible with a 1 cm safe margin was done to obtain a good prognosis. The inferior alveolar nerve was preserved and immediate reconstruction with autologous iliac crest bone graft and 2.5 mm reconstruction plates and bicortical screws was done [Figure 3].

Discussion

AF is rare neoplasm of adolescence commonly seen in the second decade of life. AF has slight predilection for males with male-to-female ratio of 1.2:1–1.4:1. The posterior mandible is the most common site of occurrence of AF (80%). Interestingly, most of the lesions in the anterior region are seen in the maxilla, and most lesions in the posterior region are seen in the mandible.[3,4,7]

AF is seldom symptomatic and usually presented as a hard swelling with intact overlying mucosa. AF exhibits slightly slower growth than simple ameloblastoma and does not infiltrate among trabeculae of bone; instead, it enlarges by gradual expansion leaving the periphery of the lesion smooth.[8] Radiographically, it resembles other odontogenic tumors. Multilocular radiolucency is most common (75%) and are seen in association with large lesions. Occasionally, it can cause root resorption of erupted teeth and bony perforation of cortical plates is also noted.[4] Similar clinical and radiographic findings were found in our case.

Microscopically, AFs are composed of neoplastic epithelial and connective tissue components. The epithelial component resembles embryonic dental lamina which consists of islands and cords of odontogenic epithelium with two or three layers of cuboidal cells and small nests or islands of cells with scanty cytoplasm while larger nests show stellate reticulum-like cells. The mesenchymal component of AF closely resembles the fibromyxoid tissue of primitive the dental papilla characterized by plump fibroblasts and delicate collagen fibrils.[8,9] If enamel or dentin is noted histologically, they are classified as ameloblastic fibro-odontoma and ameloblastic fibrodentinoma, respectively.[10]

The differential diagnosis of AF includes odontogenic cyst and tumors such as dentigerous cyst, ameloblastoma, odontogenic keratocyst, myxoma, and other mixed tumors.[8] Although these lesions show many overlapping characters, it is crucial to differentiate AF from other mixed odontogenic tumors owing to its true neoplastic potential, malignant transformation potential, and possibility of recurrence.[8,11]

Assessment of the proliferative potential of AF using immunohistochemical markers helps in understanding of tumor aggressiveness and facilitates adequate surgical planning.[12] In the present case, ameloblastic epithelium was strongly immunoreactive for CK 5 and 6/CK 14 with focal central immunopositivity in stellate reticulum for calretinin. The Ki67 proliferative index in fibromatous component was around 20% [Figure 4]. In regard with high Ki67 index, higher chances of recurrence are possible, thus long follow-up is imperative.[13]

AF has a tendency for recurrence and different recurrence rates have been reported in literature ranging from 16.3% to 33.3%.[3,4,6] The rate of malignant transformation ranges from 6.4% to 11.4% in various studies.[3,4,6] Interestingly, most of the malignant transformations are seen in older age group and most of recurrences are noted in younger age group.[14]

The appropriate treatment for AF continues to be a topic of debate. Some authors suggest treatment of smaller lesions with conservative treatment such as enucleation and curettage to limit the disability caused by more extensive procedures.

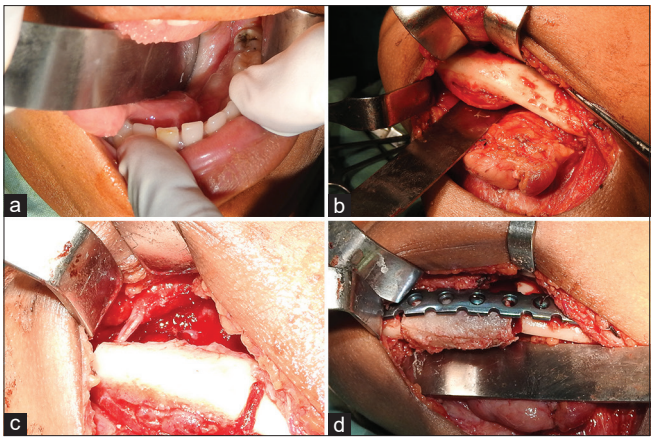

Figure 3: Surgical treatment. (a) Lingual expansion of the mandibular body in the posterior region. (b) Submandibular approach for segmental resection showing the lingual expansive lesion and loss of lower border of the mandible. (c) Osteotomy of mandible while preserving inferior alveolar nerve. (d) Immediate reconstruction of the defect using autogenous iliac crest and reconstruction plate

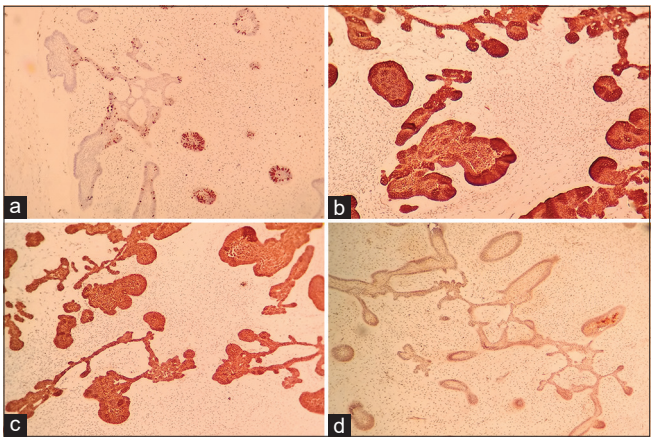

Figure 4: Immunohistochemical analysis. (a) Immunohistochemical analysis showing highly proliferative neoplastic epithelium for Ki67. (b) Immunohistochemical analysis showing strong immunoreactivity for CK 14. (c) Immunohistochemical analysis showing strong immunoreactivity for CK 5 and 6. (d) Immunohistochemical analysis showing focal central immunopositivity in stellate reticulum for calretinin

It is suggested that an extensive approach of marginal or segmental resection should be considered in patients with extensive multilocular lesions and recurrent cases.[14] In the present case, owing to large multiocular lesion in a young patient with high Ki67 index, segmental resection with safe margin of 1cm followed by immediate reconstruction with autogenous iliac crest graft was done. The follow-up of patient was done at 1 week, 3 weeks, 3 months, and thereafter at 6 months interval for 15 months, no signs of recurrence or malignant transformation were seen.

Conclusion

AF is a rare odontogenic neoplasm which shows overlapping clinical and radiographic characters with other odontogenic tumors. It relatively occurs in young age group and has potential for recurrence and malignant proliferation. IHC studies help in understanding the nature and proliferation of such tumors. Appropriate surgical modality should be followed owing to its chances of recurrence and malignant transformation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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