

CASE REPORT

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doi: 10.5866/3.4.745

Plexiform Ameloblastoma Mimicking a Periapical Cyst - Report of a Rare Case

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Article Info

Received: July 17, 2011

Review Completed: August, 19, 2011

Accepted: September, 22, 2011

Available Online: January, 2012

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ABSTRACT:

There are some rare instances of benign and malignant lesions mimicking, periapical lesions clinically and radiographically. Small incipient lesions may be mistaken for a common periapical granuloma or cyst and the tooth may be treated endodontically or extracted, with the lesion going undiagnosed leading to unavoidable complications. Therefore, histological study of any periapical lesion is mandatory in order to confirm the diagnosis and distinguish it from a non-inflammatory lesion. This report presents a plexiform variant of ameloblastoma occurring as a periapical lesion above the roots of the maxillary anterior teeth which was initially misdiagnosed as a periapical lesion and teeth were provided endodontic treatment.

Key words: Ameloblastoma, Periapical lesion, Diagnosis

INTRODUCTION

An Ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm, and only odontomas outnumber it in reported frequency of occurrence. Ameloblastoma occurs in all areas of the jaws, but the mandible is the most commonly affected area.¹ Statistics on the location of maxillary ameloblastomas are more variable and more difficult to interpret. Some studies report a low incidence in the anterior maxilla.² Presentation of this lesion as a pericoronal radiolucency associated with an impacted tooth is well documented. However the presentation of this lesion as a solitary periapical radiolucency associated with a non-impacted tooth, is rare.^{3,4} Periapical presentation of this lesion may confuse practitioners.⁴ Here we report a case which

was diagnosed initially as periapical cyst and later was confirmed as plexiform ameloblastoma histologically.

Case Report

A 40 year old male patient reported to Govt Dental College, Trivandrum with a presenting complaint of pain and swelling in upper jaw for 3 months. Patient gave a history of pain of the upper anterior teeth and subsequent root canal treatment, 3 years back. Patient was a chronic smoker with occasional alcohol intake. Intraorally the patient had a soft fluctuant swelling in relation to periapical region of 12 and 13. Palatally swelling extended from 11 to 13 region. The swelling was tender and 13 had grade I mobility. IOPA showed endodontically treated 11, 12, 13, 14 and 15. An ill defined radiolucency of size 2cm X 1.5 cm was present in relation to the apices of these endodontically treated teeth (Fig 1). All the

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teeth involved showed root resorption. Occlusal radiograph and panoramic radiograph also showed the similar picture (Fig 2). A clinical diagnosis of periapical cyst was made and lesion was enucleated along with curettage and sent for biopsy.

Gross examination of the tissue revealed multiple bits of grey coloured tissue with an aggregate size of 2.5cm X 1cm. The microscopic examination of the histological sections revealed proliferation of odontogenic epithelial islands with flattened peripheral cells, and polygonal and spindle-shaped central cells. A network of peripheral columnar pre ameloblastic cells and central loosely arranged cells resembling the stellate reticulum, could be observed in some areas (Fig 3). In addition, foci of cystic degeneration and squamous metaplasia were also identified within the epithelial nests. The tumor epithelial component was immersed in a moderately collagenous stroma. Juxta epithelial hyalinisation was evident in many areas in the connective tissue stroma (Fig 4). A definitive diagnosis of Plexiform ameloblastoma was given based on histological examination.

Discussion

Ameloblastoma is a benign but aggressive neoplasm of odontogenic origin. Ameloblastoma occurs over a broad age range; cases have been reported in children younger than 10 years through elderly adults older than 90.¹ The average age at diagnosis consistently is reported in the range of 33 to 39, and most cases cluster between ages 20 and 60 years.¹ The molar- ramus area of mandible is the most commonly affected site in the jaws. Patients with ameloblastoma most commonly present with chief complaints of swelling and facial asymmetry.

Proper diagnosis and management of ameloblastomas is not at all debatable because of the high recurrence rate demonstrated by this lesion.^{5,6,7} Recurrence rates of 50% to 90% have been associated with lesions treated by curettage alone and should by no means be underestimated. The growth characteristics of ameloblastomas vary from

other benign lesions and tumor resection is often advocated regardless of the type of ameloblastoma.¹ In spite of being classified as a benign tumor ameloblastoma is infiltrative, and locally invasive, and growth occurs in pathways of least resistance infiltrating through cancellous bone.⁶ Enlargement causes expansion and erosion of cortical bone as well as lamellar thinning. Because of this microscopic infiltration of bone, treatment also necessitates removal of some of the normal bone adjacent to the periphery of the tumour.⁵ The appropriate amount of normal bone beyond a radiographic boundary required for a tumour-free margin has not been definitively established.⁶ Marx *et al.* Advocates resection of at least 1 cm of normal-appearing bone beyond the radiographic tumor margin.⁸ Other clinicians, such as MacIntosh, recommend 2 cm or more.⁹ When the tumour has perforated bone, removal of adjacent soft tissue extending to the next adjacent anatomic boundary must be performed to ensure complete tumour-free soft tissue margins.^{3,6} Inadequate diagnosis and treatment of this lesion (ie as a periapical cyst or granuloma) although followed by initial healing, may lead to recurrence years later, but with more aggressive behavior.

Most studies about periradicular lesions have focused on periapical cysts and granulomas, ie lesions associated with pulpal necrosis and infection, owing to their high prevalence and treatment need. It is important that, after clinical and radiographic evaluation of a patient with a periapical lesion, diagnosis be confirmed by obtaining a specimen and sending it for microscopic examination (even if the lesion is small and or associated with a pulpally involved tooth) so that prompt appropriate treatment may be rendered.⁵ An incisional or excisional biopsy may be done depending on the size of the lesion and its clinical features.⁶ An incisional biopsy is recommended if a representative specimen can be obtained. This will provide the clinician with a definitive diagnosis and allow for an appropriate workup before developing a definitive therapeutic protocol.

However, non-inflammatory lesions of different nature and biological behavior mimicking inflammatory periapical disorders such as developmental odontogenic cysts, lymphomas, periapical cemento-osseous dysplasias, central giant cell lesions and ameloblastomas have been reported. Variants of ameloblastoma, are of particular concerns due to their aggressive clinical behaviour and high rates of recurrence.

In this report, a case of plexiform ameloblastoma undergoing cystic degeneration which was clinically and radiographically misdiagnosed as periapical lesion is presented. Besides, it shows the relevance of performing incisional biopsies of any periapical lesions in order to provide proper diagnosis and management. Furthermore misdiagnosis often leads to inappropriate management of the lesion, like root canal treatment (as in this case) or tooth extraction, which may further lead to progression of the original pathology and related complications

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Fig 1: Periapical Radiograph



Fig 2: Panoramic Radiograph

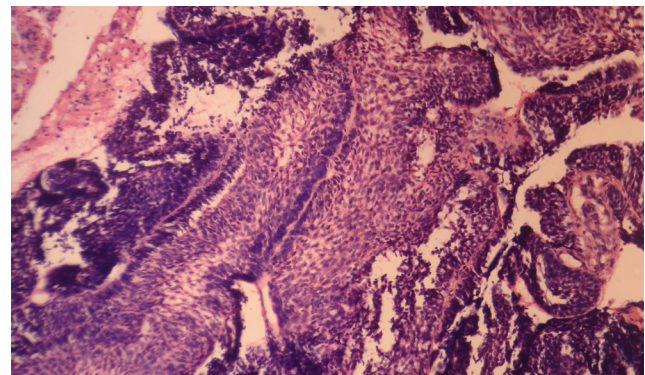


Fig 3: Photomicrograph Showing Plexiform Pattern

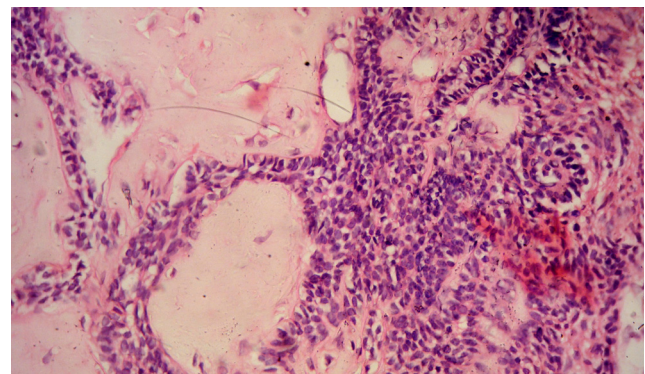


Fig 4: Photomicrograph Showing Juxtaepithelial Hyalinisation