



Plexiform Ameloblastoma with Acanthomatous Changes: A Rare Case Report

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Abstract

Ameloblastoma, One of the most Common and Frequently Encountered Odontogenic Epithelial Derived Tumours of the oral Cavity, are Reported to be Represented by Variable Number of Clinical, Radiological and Histological Types. Also, this Lesion has been Observed to be most Aggressive and has a Tendency to Recur in most Circumstances. The Histological Variants of Solid type of Ameloblastoma have also been Observed to Occur either in Isolation or in Combination with One or the Other Lesions. Hence, this case Report is One of those Combined Lesions Presenting with Little Clinical Presentation, but Histopathologically was Diagnosed to be Plexiform Ameloblastoma with Acanthomatous Changes. These Types of Lesions have been Scarcely Reported in Literature and should be Reported with Regular follow-up so as to not only Improve the Diagnostic Criteria, but also to Improve Surgical Outcomes, Making the Patients Prone to less Morbid Outcomes.

Keywords Odontogenic tumours · Ameloblastoma · Posterior mandible

Introduction

Odontogenic tumours can be variably defined as tumours or tumour-like growth that have their origin from odontogenic tissues, the heterogeneity amongst them being present as a result of varying induction that occurs between developing embryonic components which under normal conditions, would lead to the formation a normal tooth germ [1]. One such lesion is Ameloblastoma, which is the most common odontogenic tumours encountered in the jaws (48.9%) and represents 9% of all the odontogenic epithelial lesions found in the jaws, which itself causes minimal inductive changes in the connective tissue. It is a true neoplasm of enamel organ with certain peculiarities such as persistent local growth, high loco-regional invasion, non-encapsulated in nature and rarely metastasizing which does not undergo differentiation till the point it is capable of forming hard tissues(enamel). It was first coined as adamantinoma by Broca in 1868 and then defined by Malassez in 1885,

who suggested that it may arise as a result of remnants of dental lamina [2]. It was then described by Robinson in 1937 as a benign tumour that is —usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent. The World Health Organization (1991) defined ameloblastoma as a benign but locally aggressive tumour with a high tendency to recur, consisting of proliferating odontogenic epithelium lying in a fibrous stroma [3]. In 2005 WHO, had categorized ameloblastoma into (1) peripheral, which comprises an extraosseous counterpart and shows continuity with the oral mucosal stratified squamous epithelium, (2) unicystic, consisting of single cystic intraosseous growth pattern (3) solid/multicystic, characterized by invasive tumour which penetrates bone marrow spaces and may show multicystic foci (4) desmoplastic, comprising of an infiltrative intraosseous tumour dominated by the stromal component, radiographically reminiscent of a fibro-osseous lesion [4]. However, desmoplastic ameloblastoma was re-included only as a histological subtype in the 2017 WHO classification [5]. The tumour most commonly is found in jaw bones and has the potential to erode through the cancellous bone and it has been thought to arise through one of the following sources: - (1) Cell rests of the enamel organ, (2) epithelium of the odontogenic cysts, (3) disturbances of the developing enamel organ, (4) basal cells of the

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surface epithelium and (5) heterotrophic epithelium in other parts of the body [6]. It has variable clinical, radiological and histological characteristics, with a tendency to recur, if not excised in toto in a single go. The aetiology is unknown, but many associated factors have been proposed including nonspecific nonirritating factors such as extraction, caries, trauma, infection during tooth eruption or viral pathogenesis [7]. The lesion has most common tendency to occur in the mandibular molar-ramus region and is most often asymptomatic in its presentation [8]. It can occur in any decade of life, but most commonly presents in third to fourth decade, and may present clinically as swelling, loosening of teeth, malocclusion and sometimes pain [9]. The radiographic features are variable, ranging from multilocular radiolucency with scalloped cortical margins resulting in “soap-bubble appearance” to either expansion or resorption of cortical plate resulting in displacement, root resorption and loss of the teeth in the associated region [10]. Histologically, the variants of multicystic/ intraosseous ameloblastoma have been categorized as Follicular, Plexiform, Granular, Acanthomatous, Basaloid and Desmoplastic Ameloblastoma, occurring alone or in combination with one another [11]. The diagnostic criteria for identification of ameloblastoma was given by Vicker’s and Gorlin in 1970, which included: - hyperchromatism of basal cell nuclei, basal cell palisading with polarization which is referred to as reversal of polarity and cytoplasmic vacuolization with intercellular spacing of lining epithelium [12]. Management of ameloblastoma poses a challenge for all of those practitioners involved in the field of head and neck surgery because successful treatment requires not only adequate resection but also the functional and aesthetically acceptable reconstruction of the residual defect. Resection with wide margins and reconstruction in the same sitting is currently accepted as



Fig. 1 Showing the radiographic appearance of the lesion

the treatment of choice in most cases. Idea of conservative surgery is no longer entertained since it is associated with higher recurrence rate [13].

The following study focuses on a case report which has reported to the department with only a complaint of swelling in the left posterior mandible with no associated symptoms which was then diagnosed to be Plexiform Ameloblastoma with Acanthomatous changes, which has been rarely reported in Literature.

Case Perspective

A patient reported to the department with the chief complaint of swelling in left lower back region of the jaw for 1 year. The swelling was gradual in onset, which slowly progressed to the size at the time patient reported. The swelling was not associated with any other symptoms of pain, induration or discharge from the affected region. Patient upon questioning further, did not revealed any history of substance abuse. Patient had a history of being diagnosed with Diabetes Mellitus in the last 2 years and is currently not on medication for the same, also had falling off the teeth in the region of swelling without any sort of intervention. Upon extraoral clinical examination, no gross facial asymmetry was noted. On intraoral clinical examination, hard, bony, painless swelling was noted extending from distal aspect of 35 without any extension into the soft tissues or extruding to the mucobuccal fold, being completely localised in the alveolar bone, and extended till the retromolar trigone region. Tooth corresponding to the same site viz., 36, 37, 38 were noted to be missing. Palpation confirmed the clinical findings. After the clinical examination, patient was sent for Cone- beam Computed tomography imaging. The imaging revealed a well demarcated, multilocular predominantly radiolucent lesion seen extending from the left side of the mandibular body region till the ramal region (Fig. 1). Then the patient was sent for haematological examination which revealed the fluid proteins level to be in higher range (987.50 mg/ dl: Normal range – 150–450 mg/dl). The provisional diagnosis of Odontogenic Keratocyst was given. Patient underwent complete surgical excision of the lesion and it was the fixed in Formalin and sent for histopathological examination. (Fig. 2).

Tissue underwent routine processing and then was stained with Haematoxylin and Eosin and the slides obtained were examined under examination. The examination revealed odontogenic islands arranged in the form of anastomosing pattern bounded by tall columnar cells with hyperchromatic nucleus in a palisaded arrangement showing reversal of polarity and subnuclear vacuolization. The central core of the section revealed loosely arranged stellate reticulum like

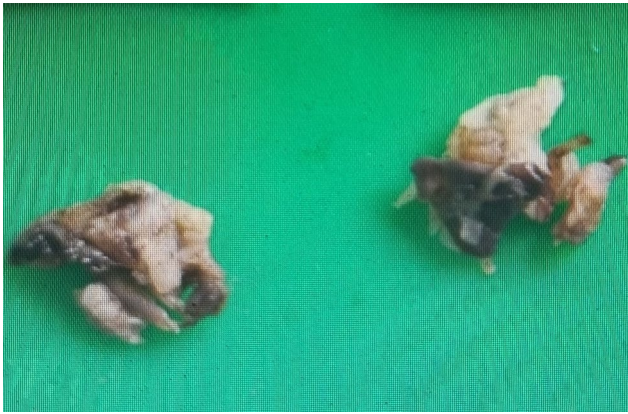


Fig. 2 Showing the complete excised tissue from the site

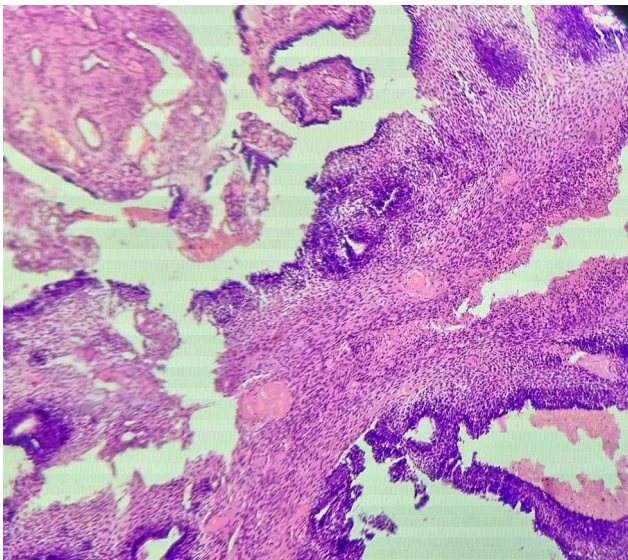


Fig. 3 Showing the histopathology at 4x

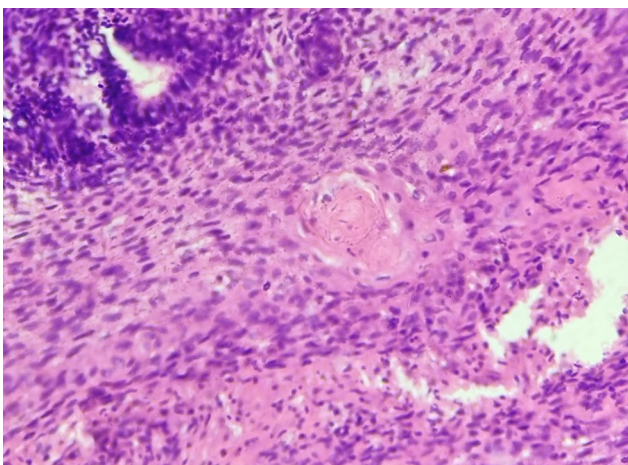


Fig. 4 Showing histopathology at 40x

areas with evidence of cystic degeneration and squamous differentiation. The histopathological features confirmed the diagnosis of Plexiform Ameloblastoma with Acanthomatous changes (Figs. 3 and 4).

Patient has been put on regular follow-up after surgical excision and reported no signs of recurrence after 6 months of excision.

Discussion

Plexiform ameloblastoma most commonly occurs during fifth to sixth decade of life. Most common site of occurrence is the posterior mandible. Plexiform Ameloblastoma has been classified histopathologically to be composed of cells arranged in interconnecting strands and cords with cuboidal or columnar basal cells exhibiting hyperchromatic nuclei, nuclear palisading with polarization and central stellate reticulumlike cells [14]. Acanthomatous ameloblastoma most common occurs during seventh decade of life [15]. The most common site of occurrence is the posterior mandible (80% of the cases). Histopathologically, acanthomatous type shows central squamous cell differential with keratin formation as seen in the present case. Some authors stated that formation of squamous metaplasia may be due to chronic irritation of calculus and oral sepsis [16]. Waldron and El-Mofty discovered a rare variant of ameloblastoma composed histologically of areas of the classic follicular or plexiform ameloblastoma and areas of desmoplastic ameloblastoma. This was named hybrid ameloblastoma (HA). Till 2016, > 30 cases with hybrid ameloblastoma have been reported in literature till date [17]. Hybrid ameloblastoma has a variable clinical, radiological, and histopathological presentation. They contain two or more different histologic types and their biologic compartment is still arguable. Mandible is the most common site for involvement of hybrid ameloblastomas as compared to the maxilla [18]. Radiographically, it mimics fibro-osseous diseases and odontogenic cysts and tumours with mixed radiolucent-radiopaque internal structure such as ossifying fibromas, fibrous dysplasia, osteoblastomas, osteosarcomas, calcifying epithelial odontogenic tumours, and calcifying odontogenic cysts [19]. With limited understanding of its behaviour and prognosis, the proper treatment strategies for hybrid ameloblastoma are not entirely defined so far. Based on the present knowledge, the WHO recommends to apply the same treatment modality as for solid ameloblastoma which includes complete resection as enucleation or curettage might result in its recurrence. However, small lesions can easily be enucleated in toto [20]. This kind of hybrid lesions is first amongst of its kind to be reported in the literature.

Conclusion

Hybrid ameloblastoma has a variable clinical, radiological, and histopathological presentation. The biologic comportment of the lesion is still arguable. Immunohistochemical markers may act as an adjunct in the accurate diagnosis of this lesion. Thus, many more clinical, radiological, and histopathological data are required to clearly demonstrate this pathologic entity. For this to take place, many case reports having such hybrid features should be increasingly reported so as to work out on all the arenas surrounding these lesions. The lesion reported in this article is Plexiform Ameloblastoma with Acanthomatous changes which has scarcely been reported in literature till date. So, a greater number of cases are required to form a basic sound literature proof.

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