Recurrence of Plexiform Ameloblastoma as Acanthomatous Ameloblastoma: A Rare Case Report

Abstract

Ameloblastoma is an uncommon locally invasive benign odontogenic tumor arising from the odontogenic epithelium. It is a slow-growing tumor with locally aggressive nature, and posterior mandible is the most common location. The recurrence rate is high even after *en bloc* resection. Acanthomatous ameloblastoma is one of the rare variants of ameloblastoma and is usually seen in older age group. Here, we present a case of acanthomatous ameloblastoma in a 46-year-old female who previously diagnosed and treated for a case of plexiform ameloblastoma 17 years back. This is the first case to be reported among ameloblastoma with different histopathological variants at recurrence.

Keywords: *Acanthomatous, ameloblastoma, plexiform, recurrence, survival*

Sanat Kumar Bhuyan, Ruchi Bhuyan[1], Tapan Kumar Sahoo[2], Pinali Das

Departments of Oral Medicine and Radiology and [1]Oral Pathology and Microbiology, Institute of Dental Science SOA University, Bhubaneswar, [2]Department of Radiotherapy, HCG Panda Cancer Hospital, Cuttack, Odisha, India

Introduction

Ameloblastoma is a benign odontogenic tumor arising from the odontogenic epithelium and is locally invasive. Ameloblastoma is reported to constitute about 1% of all the tumors and cysts of jaw and 18% of odontogenic tumors.[1,2]

Cell rests of enamel organ, epithelium of odontogenic cysts, disturbances of developing enamel organ, basal cells of surface epithelium and heterotopic epithelium in other parts of the body are the possible sources of epithelial origin of ameloblastoma.[3] On the basis of histopathological classifications, ameloblastoma is of six types such as follicular, plexiform, desmoplastic, acanthomatous, basal cells, and granular cell. Papilliferous, clear cell, and keratoameloblastoma are the other possible rare verities.[4,5] Follicular ameloblastoma is the most common histological variant (64.9%), followed by the plexiform (13.0%), desmoplastic (5.2%), and acanthomatous (3.9%) varieties.[3] These variants may exist singly or multiple. According to the current World Health Organization classification, ameloblastoma can be classified as solid/multicystic type, extraosseous/peripheral type, desmoplastic type, and unicystic type.[1]

Biological behavior and the rate of recurrence are different in these tumor types requiring different forms of treatment. The treatment of ameloblastoma can be conservative or radical approach and depends on the type, location, and size of the lesion and age of the patient.[6]

Here, we report a case of recurrent ameloblastoma of the left side mandible as multicystic acanthomatous variant in a 46-year-old female after 17 years of treatment for plexiform ameloblastoma at the same location.

Case Report

A 46-year-old female presented with a chief complaint of swelling in the left lower jaw toward the posterior aspect for 3 months, and the swelling was gradually increased in size to the present size with intermittent pain. On extraoral examination, a diffuse swelling was present on the left side of the face extending from the angle of mandible posteriorly to ramus superiorly and posterior submandibular region inferiorly measuring about 5 cm × 4 cm in size. Overlying skin was normal with no evidence of discharge [Figure 1a]. On palpation, swelling was firm, mild tender, nonpulsatile, and noncompressible. Intraoral examination revealed a diffuse swelling in the left retromolar region and buccal vestibule in 33–38 region [Figure 1b]. There

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Address for correspondence:
Dr. Tapan Kumar Sahoo, Department of Radiotherapy, HCG Panda Cancer Hospital, Cuttack, Odisha, India.
E-mail: drtapankumars8@gmail.com

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was absence of any significant cervical lymph node on palpation. The patient had a history of swelling in the same location 17 years back for which she underwent surgical excision and diagnosed as plexiform ameloblastoma on histopathological examination [Figure 2a-c].

Radiograph of the mandible revealed expansion of buccal and lingual cortical plates with multilocular, soap bubble appearance [Figure 3a]. Noncontrast computed tomography (CT) and three-dimensional CT scan revealed an expansile lytic multicystic lesion of size 5.5 cm × 3.0 cm × 2.7 cm with well-demarcated margin in the body of the left hemimandible with air foci within it and the lesion not crossing the midline [Figure 3b and c]. The mass was abutting the left margin of the tongue, left mylohyoid, and buccinator muscle. With the above radiological features, the patient was suspected as a case of recurrent ameloblastoma.

Fine-needle aspiration cytology from the lesion showed blood-tinged, straw-colored fluid with inconclusive diagnosis. Hemimandibulectomy was performed with sufficient, safe margins under the general anesthesia. Multicystic appearance of the lesion was found on intraoperative finding [Figure 4].

Histopathological examination of the operated specimen revealed delicate collagenous stroma with numerous odontogenic follicles comprised peripheral ameloblast like-cells and central stellate reticulum-like cells. Multiple follicles were showing squamous metaplasia of stellate reticulum-like cells, some of which were transferred to keratin pearls. Many follicles also showed evidence of cystic degeneration. Diffusely arranged mixed chronic inflammatory cells were present in vascular stroma. With these features, the final diagnosis of acanthomatous ameloblastoma was confirmed [Figure 5a-c]. The patient was doing well without any recurrence during 6 months of follow-up.

Discussion

Ameloblastoma is a locally aggressive, benign tumor of the oral cavity and is the second most common odontogenic tumor, the first being odontoma. Majority of cases are asymptomatic and detected on routine dental X-rays. A slow-growing, painless swelling or expansion of the jaw resulting facial asymmetry is the common presentation, and malocclusion of teeth may occur. The lesion may erode through the bone and extend into the soft tissues.[4] Majority of ameloblastomas present in third–fifth decades of life with equal sex predilection. Whereas, acanthomatous variants commonly present in the seventh decade of life.[1,7] Mandible is the most common site of presentation for ameloblastoma consisting of approximately 80% of cases, with majority being located in posterior aspect of the mandible, i.e., molar or ascending ramus area and 10%–15% of ameloblastomas may be associated with a nonerupted tooth.[4]

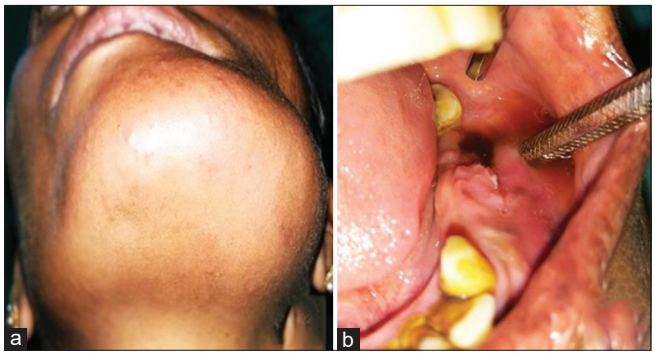

Figure 1: (a) Swelling over the left side neck on extraoral view and (b) swelling in the left side retromolar region on intraoral view

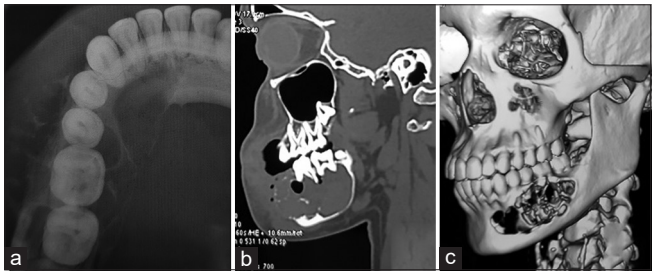

Figure 3: Occlusal radiograph of the left side mandible showing soap bubble appearance (a), and an expansile lytic multicystic lesion of size 5.5 cm × 3.0 cm × 2.7 cm with well-demarcated margin in the body of the left hemimandible on noncontrast computed tomography (b) and three-dimensional computed tomography scan (c) of the face and neck

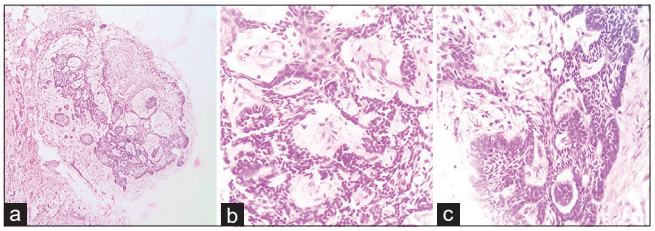

Figure 2: (a-c) Section from the initial biopsy showing the anastomosing strands of basal cell, inconspicuous reticulum, and delicate stroma. (H and E, a: ×40, b and c: ×400)

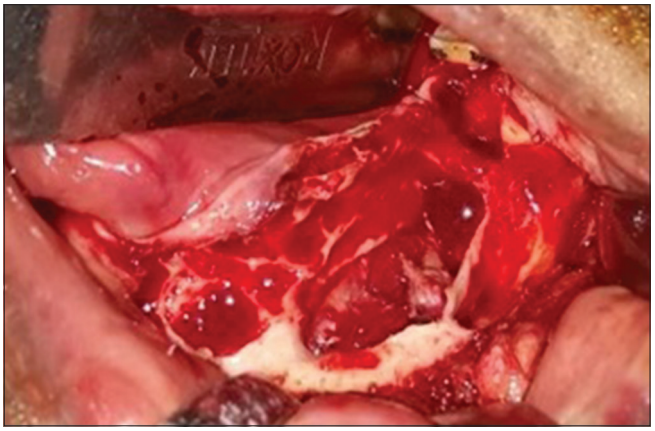

Figure 4: Multicystic appearance of the lesion on intraoperative finding

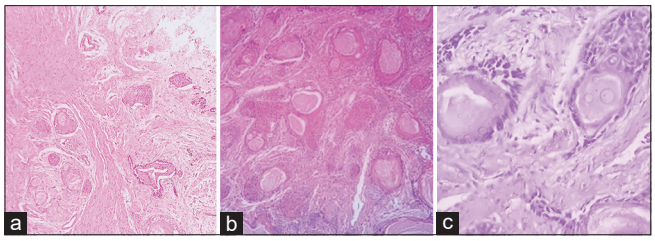

Figure 5: Section from the present biopsy showing (a) nests of odontogenic follicles having surrounding fibrous stroma, (b and c) the presence of multiple odontogenic follicles containing peripheral ameloblast-like cells and central stellate reticulum cells and follicles showing squamous metaplasia. (H and E, a: ×40, b: ×100, c: ×400)

Ameloblastoma on radiograph commonly shows a radiolucent, unilocular/multiloculated cystic lesion, with a characteristic "soap bubble-like" appearance, cortical thinning/destruction, local invasion, and root resorptions.[8] CT scan is the most promising in identifying cortical destruction and soft-tissue involvement due to infiltration of the tumor cells predominantly into cancellous portion of the cortical bone.[6]

Microscopically, ameloblastomas have a fibrous stroma with islands or masses of proliferating epithelium resembling odontogenic epithelium of the enamel organ and palisading of cells around proliferating epithelium in a pattern similar to ameloblasts. The follicular pattern consists of islands of epithelium containing a core of loosely arranged angular cells resembling stellate reticulum, surrounded by a single layer of tall columnar cells with reverse polarity of the nuclei. The plexiform pattern shows long anastomosing cords or sheets of odontogenic epithelium.[5] On histopathological examination, acanthomatous ameloblastoma shows central squamous cell differential with keratin formation supporting our findings. Such squamous metaplasia may be due to chronic irritation of calculus and oral sepsis.[1]

The existence of multiple histopathological patterns in the same tumor at initial diagnosis of ameloblastoma has been reported in few cases only. For example, Jain *et al*. reported the presence of luminal and mural types with acanthomatous and granular cell changes in a case of unicystic ameloblastoma.[9] However, the presence of different type of histopathological pattern at the time recurrence of ameloblastoma is quite rare. Hertog *et al*. in a study found that two patients with an initial plexiform and mixed type ameloblastoma, after treatment with enucleatiion develop recurrence with different histopathological types from the primary tumor, i.e., desmoplastic variant.[10] After extensive search of literature, there is no case reported with changing histopathological pattern from plexiform type to acanthomatous type during recurrence of ameloblastoma. Therefore, our case is unique one to be reported first time with such different histopathological type during recurrence. The possible explanation of such changing histopathology type in our case is the risk of that area for chronic irritation due to calculus or oral sepsis resulting squamous metaplasia and changing histopathology from plexiform to acanthomatous variant during recurrence.[1]

Ameloblastoma has potential for local invasion and tendency to recur. The treatment approach for ameloblastoma is either conservative or radical approach. Enucleation is the common conservative approach and particularly use for unicystic type. Whereas, radical approach is commonly used for large ameloblastoma involving the inferior alveolar canal or below or for more aggressive variants such as intramural ameloblastoma or multicystic type. Segmental or marginal resection with 1.5–2 cm normal bony margin beyond the radiologic margin is used in radical approach of ameloblastomas.[6] In addition to low sensitivity of this neoplasm, the intraosseous location of the ameloblastoma prevents the use of radiotherapy as an effective therapeutic option because radiation leads to osteonecrosis and induces the potential development of secondary tumors. Therefore, in all types of ameloblastomas, a thorough long-term clinical and radiographic follow-up is always recommended.[4]

The recurrence rate of ameloblastoma is 13%–15% for surgical resection, whereas it was 90%–100% for curettage.[7] The recurrence rate is high with mandibular ameloblastomas and in follicular histological type than in maxillary ameloblastomas and in plexiform or other histological variants, respectively.[8] Unicystic ameloblastoma has a fibrous connective tissue capsule and therefore has a much lower rate of recurrence. The solid or multicystic ameloblastoma has a tendency to be locally invasive and with a recurrence of 90% after conservative management such as curettage and enucleation.[11] Recurrence of the lesion is especially due to infiltrative growth of tumor into the cancellous bone. Also, periosteum invasion of the tumor may involve soft tissue component resulting inadequate resection and may leads to recurrence. The chance of recurrences for ameloblastoma presents within the first 5 years, whereas, in our case, recurrence occurs after 17 years of follow-up.[8] Some authors observed recurrence in the patients >10 years after the initial treatment.[12]

In summary, acanthomatous ameloblastoma is a rare entity, and the present case is unique one due to recurrence of such variant at the same location with a different histopathological pattern, i.e., from the initial plexiform pattern to the present acanthomatous pattern after a long duration of follow-up of 17 years. To our knowledge, this is the first case to be reported with such type of different histopathological pattern during recurrence and this helps the clinician for treatment and further exposure of such possibilities and outcome.

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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