Case Report

Ameloblastic carcinoma: A case report and literature review

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Abstract

Ameloblastic carcinoma (AC) is a rare epithelial odontogenic tumor of the jaws, which exhibits cytological features of ameloblastoma and carcinoma. It has a distinct predilection for mandible. These lesions may initially show histologic features of ameloblastoma that dedifferentiate over time. Others may present with features of epithelial dedifferentiation in ameloblastoma. A case of ameloblastomic carcinoma in a 64-year-old male is reported, who presented with swelling in the left mandible 3 months after the extraction of the left upper molar.

Key words: Ameloblastoma, carcinoma, odontogenic

Introduction

Ameloblastoma is a benign but locally aggressive neoplasm which clinically presents as a slowly growing painless swelling of the jaw. Eighty percent of it arises in the mandible.[1] It may present as a cystic lesion with benign clinical features or as a large tissue mass with ulceration, significant bone resorption and tooth mobility.[2] The typical ameloblastoma may show a variety of histologic patterns, namely, follicular, plexiform, acanthomatous, basal cell, and granular cell types. These patterns do not have a distinct bearing on clinical behavior or prognosis, and more than one morphologic pattern may be present in a given tumor. Although the typical ameloblastoma is a histologically benign and slowly growing tumor, it has the capacity to cause destructive local growth and even death by invasion of vital structures.[1]

Rare variants are malignant ameloblastoma and ameloblastic carcinoma (AC). The classification system by World Health Organization (WHO) defines malignant ameloblastoma as an ameloblastoma, which has metastasized but exhibits the well-differentiated morphologic features of a typical ameloblastoma in both the primary and metastatic sites.[1] In comparison, AC is the pathologic designation describing an ameloblastoma with areas of obvious histologic malignancy. AC may arise *de novo* or from transformation of a long-standing primarily benign lesion, usually secondary to recurrences associated with multiple surgical procedures or radiation therapy.[1] The incidence of AC is greater than that of malignant ameloblastoma by a 2:1 ratio and carries a bad prognosis.[2]

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Case Report

A 64-year-old male presented with toothache since 4 months in the left upper molar, following which tooth extraction was done. This resulted in failure of healing of the extraction socket and a soft tissue growth over it 3 months post extraction, causing a left mandibular swelling. Incisional biopsy was done and a diagnosis of ameloblastoma was made following which surgical excision of the tumor was done. The patient presented with recurrence of the swelling after 2 years. Clinical examination revealed a diffuse, non-tender swelling of the buccal sulcus of the mandible along with an intraoral sinus. The skin overlying the swelling seemed normal. No lymph nodes were palpable on neck examination. Radiological examination of head and neck revealed radiolucent lesion involving the body and ramus of the mandible, with ill-defined margins, along with destruction of the cortical plate. Chest X-ray and further clinical examination failed to reveal any metastasis. Incisional biopsy was done from the lesion.

On microscopic examination, the neoplasm was observed to be containing follicles and cords of cohesive, poorly differentiated malignant cells with a basaloid appearance [Figure 1]. The cells displayed pleomorphism with hyperchromatic nuclei with nucleoli and scant cytoplasm [Figures 2 and 3]. Mitosis was frequent. No stellate reticulum or squamous metaplasia was identified. Thus, a diagnosis of AC was made.

Discussion

Ameloblastomas constitute a group of particularly interesting lesions because they display origin from embryologic components of the developing tooth germ. Although very rare when viewed in the context of the human tumor pathology, ameloblastoma represents the most common tumor derived from tooth epithelial components, representing between 13% and 24% of the odontogenic neoplasms.[3]

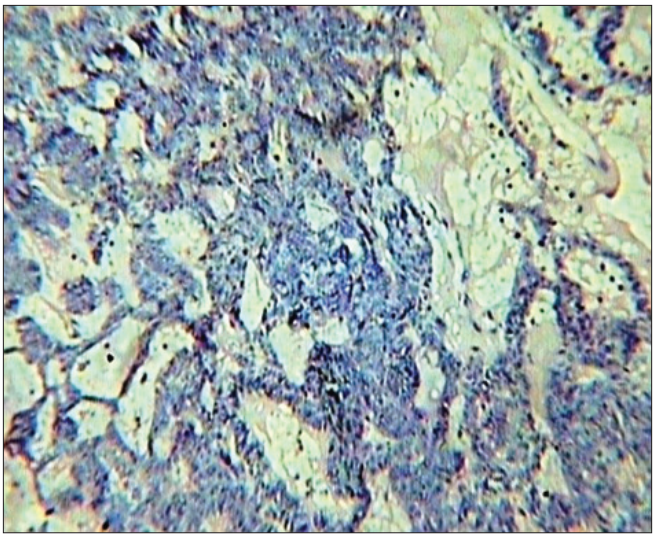

Figure 1: Low-power view showing the follicular arrangement of cells and the overall basophilic staining quality (H&E, ×100)

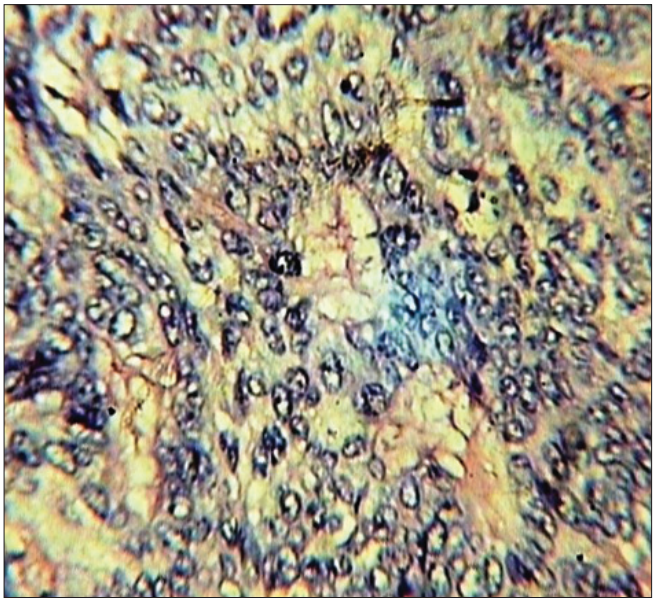

Figure 2: High power of the follicle and pleomorphic cells with prominent nucleoli (H&E, ×400)

Malignant epithelial odontogenic tumors, which include malignant ameloblastoma, AC, primary intraosseous squamous cell carcinoma, clear cell odontogenic tumor and malignant epithelial ghost cell tumor, are very rare.[4]

In 1983, Shafer introduced the term "ameloblastic carcinoma" to describe ameloblastomas in which there had been histologic malignant transformation.[2] Carcinomas associated with ameloblastoma have had several terminologies within the medical literature, thus posing a problem in accurately separating malignant ameloblastoma from AC. Several authors have attempted to make a distinction between these two entities because AC is clinically more aggressive. These definitions include a well-differentiated ameloblastoma with histologically malignant epithelial component; a tumor with histologic evidence of malignancy and features of ameloblastoma and concomitant squamous cell carcinoma; a tumor with combined features of an ameloblastoma with less differentiated areas; and any ameloblastoma with histologic evidence of malignancy in the primary tumor or the recurrent tumor, irrespective of whether the tumor has metastasized.[1]

AC is a very rare tumor with an incidence of less than 1%. It occurs in a wide range of age groups with a mean age of 30.1 years. There is no apparent sex predilection. The most commonly involved area is the posterior portion of the mandible.[2] Unusual locations are maxilla, nasal cavity and anterior skull.[5,6] Our patient was 64 years old and presented with swelling of the left mandible. The most common signs

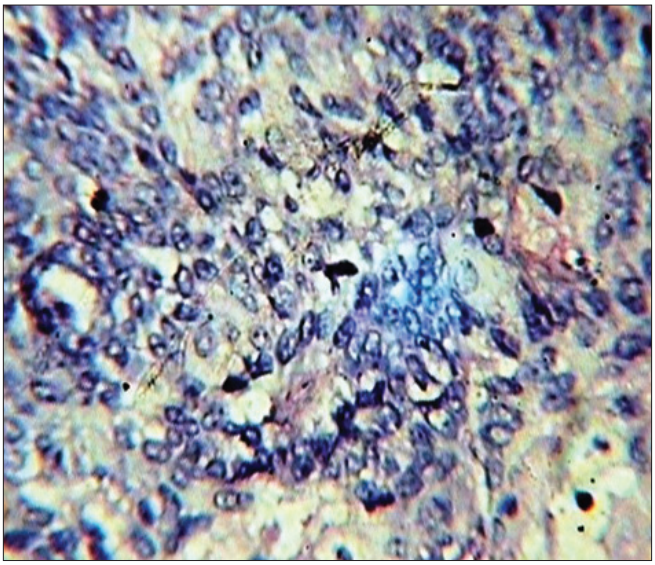

Figure 3: High-power view showing pleomorphic cells and abundant mitoses (H&E, ×400)

include swelling, associated pain, rapid growth, trismus and dysphonia. Perforation of the cortical plate, extension into surrounding soft tissue, numerous recurrent lesions and metastasis, usually to cervical lymph nodes, can be associated with AC.[2]

The radiographic appearance of the lesion is usually similar to that of ameloblastoma except for the presence of focal radiopacity, apparently reflecting dystrophic calcification.[2,7] The case described in this report also showed similar radiological features with destruction of the cortical plate.

Histologically, the tumor cells resemble the cells seen in ameloblastoma but show cytologic atypia, cellular pleomorphism, nuclear hyperchromasia, mitoses, vascular and neural invasion.[7] Similar features were noted in this case.

AC can recur locally 0.5–11 years after definitive therapy. Distant metastasis may appear 4–12 years postoperatively and is common in lung, bone, liver and brain.[6]

Chromosomal imbalances in ameloblastomas are reported to be rare, with losses in chromosomes 22 and 10 being most frequent. Aneuploidy is more common in AC and may predict malignant potential.[3]

Differential diagnosis of AC includes primary intra-alveolar epidermoid carcinoma, acanthomatous ameloblastoma, keratoameloblastoma, squamous odontogenic tumor and metastatic tumors from lung, breast and gastrointestinal tract.[2]

To conclude, ameloblastoma shows a variety of histologic and biologic behavior ranging from benign to frank malignancy. Although AC is rare, it is important to rule it out in patients presenting with toothache or mobile teeth in association with persistent jaw swelling, pain and rapid growth through prompt radiological and histopathologic investigations. Cases of ameloblastomas should thus be studied carefully to detect subtle changes in histology that may predict aggressive behavior and bad prognosis.

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