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Atypical and Varied Presentations of Ameloblastoma: A Case Series

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ABSTRACT

Ameloblastoma is a benign odontogenic tumor that exhibits local aggressiveness. Around 80% cases occur in the mandible, out of which the majority of cases are seen in the mandibular ramus region. Ameloblastoma presents as a non-symptomatic lesion and may be detected during routine radiographic examination. However, the diagnosis is confirmed by histopathological examination. In our case series, we report three cases of Ameloblastoma: one in the maxilla and two in the mandible, of which one was in the ramus region and the other in the anterior region. All lesions were confirmed by histopathological examination with immunohistochemical staining. Histologically, Ameloblastoma has multiple microscopic variants. In our study, we found a follicular variant in the maxilla, a desmoplastic variant in the mandibular ramus, and a follicular and plexiform (mixed) pattern in the anterior mandible. In all three cases, there were varied clinical and radiographic findings. Thus, histopathological examination is a crucial confirmatory tool for accurate diagnosis and effective treatment planning.

1. Introduction

Ameloblastoma is the second most common benign odontogenic tumor with locally aggressive behavior. It is also the most common tumor to be confirmed histopathologically.[1] It originates from odontogenic epithelium, such as cell rests of Malassez or cell rests of Serre, heterotopic epithelium in the pituitary gland, odontogenic cysts (such as dentigerous cysts), odontomas, and basal cells of the surface epithelium of the jaws.[2] In 2017, the World Health Organization (WHO) classified Ameloblastoma into four sub-categories: conventional Ameloblastoma, unicystic Ameloblastoma, peripheral or extraosseous Ameloblastoma, and metastasizing Ameloblastoma.[3] The primary underlying cause remains uncertain, but it may be related to localized trauma, nutritional deficiency, mutations, inflammation, or molecular aberrations impacting signaling pathways.[4] The genetic and molecular aberrations add aggressive characteristics and metastatic possibilities to the tumor. Ameloblastoma is most commonly located in the mandibular body and ramus region and is observed in individuals aged 30 to 50 years with no sex predilection.[5] However, fewer cases are reported in children.[6, 7] It is seen as pain-free non-symptomatic bony swelling which slowly progresses by cortical expansion, perforation, and infiltration into surrounding soft tissue.[8] Radiographic examination is marked by unilocular or multilocular radiolucency having a "soap bubble" or "honeycomb" shape.[5] In this case series, we report three male patients who were diagnosed with Ameloblastoma through histopathological examination. The first case exhibited a follicular pattern in the maxilla, the second case showed a desmoplastic pattern in the mandibular ramus, and the third case displayed a follicular and plexiform (mixed) pattern in the mandibular anterior region. All three cases were confirmed using immunohistochemistry.

2. Case Presentation

Case 1

A 47-year-old male patient had a history of upper right side lip swelling for 4 years. On examination, a 3x3 cm² non-tender bony hard swelling extending from the right side of the upper gums to the right side of the hard palate. CT scan of the PNS showed an expansile lytic lesion measuring 4 × 3.3 × 3.4 cm. It involved the right anterior maxillary alveolus in relation to the right first to fifth teeth and the left first tooth. It also exhibited areas of internal septation, the floor of the nasal cavity, the nasal septum, and the hard palate. It was indenting on the upper lip and the inferior turbinate. No intrasinus extension was seen. Radiological findings were suggestive of a benign etiology, most probably giant cell granuloma. In the pathology department, we received an excised specimen of bony tissue from the right maxillary alveolar area. Microscopically, it revealed islands of odontogenic epithelial cells, accompanied by peripheral palisades and stellate reticulum-like cells (Fig. 1). Focal sites of cyst formation and reactive bone formation were also observed. A diagnosis of Ameloblastoma with a follicular pattern was suggested. Immunohistochemically, tumor cells were positive for CD138 and CK19.

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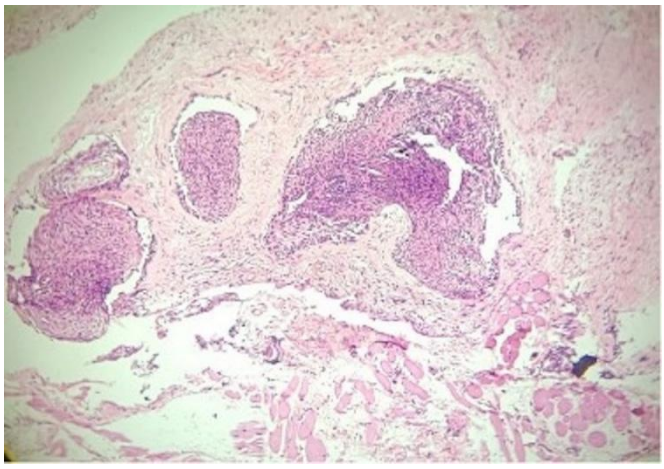

Fig. 1. Islands of odontogenic epithelial cells with peripheral palisades and stellate reticulum-like cells.

Case 2

A 32-year-old male patient had a history of swelling over the left side of his jaw for one and a half years. Upon examination, it was a bony, hard, non-tender swelling. Radiological examination revealed a cystic lesion measuring 30.5mm x 16.4mm. Clinical diagnosis was a residual cyst. In the Pathology Department, we received multiple soft tissue samples that were cream-colored and measured 1.5 cm x 0.5 cm in size. Microscopically (desmoplastic) dense cellular fibroconnective tissue and collagenous stroma within compressed and angular islands of epithelial tumor cells were observed. (Fig. 2). Immunohistochemically, the tumor cells were positive for CD138 and negative for CK19.

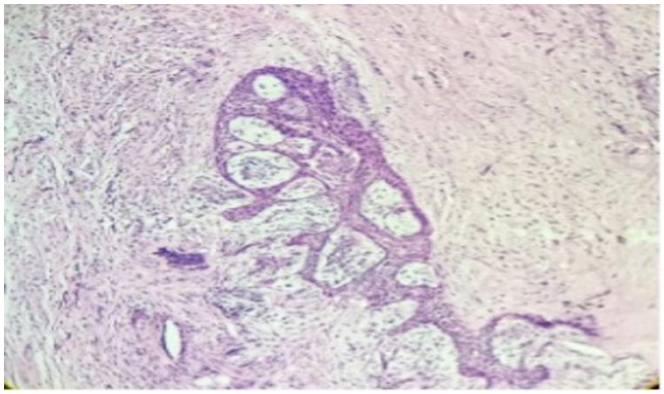

Fig. 2. Islands of neoplastic ameloblastic epithelium surrounded by desmoplastic stroma.

Case 3

A 52-year-old male patient had a history of pain and swelling in the lower anterior mandibular region for 6 months. On radiological examination, there was a cystic expansile lesion involving the anterior mandibular alveolus in relation to the right first to fourth teeth. In the pathology department, we received multiple soft tissue samples that were cream-colored and measured 2.5x1.5x0.5cm. Microscopic examination revealed islands and interdigitating cords of odontogenic epithelium, along with peripheral palisades and stellate reticulum-like areas, within fibrous connective tissue. (Fig. 3). The findings were suggestive of a follicular and plexiform (mixed) pattern in Ameloblastoma. Immunohistochemically, the cells were positive for CD138 (Fig. 4) and CK19 (Fig. 5).

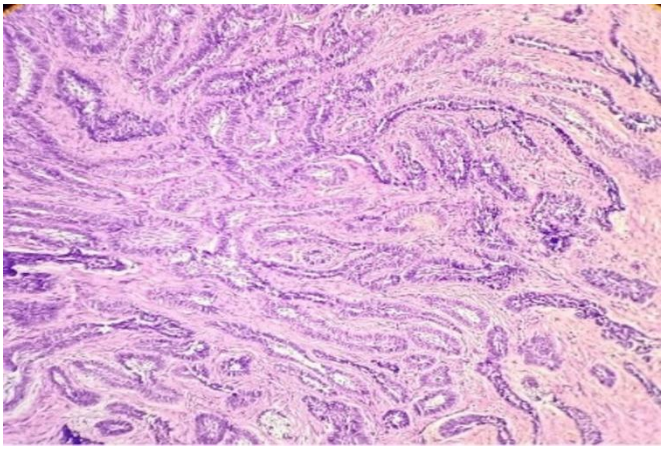

Fig. 3. Ameloblastoma tumor cells showing a plexiform pattern.

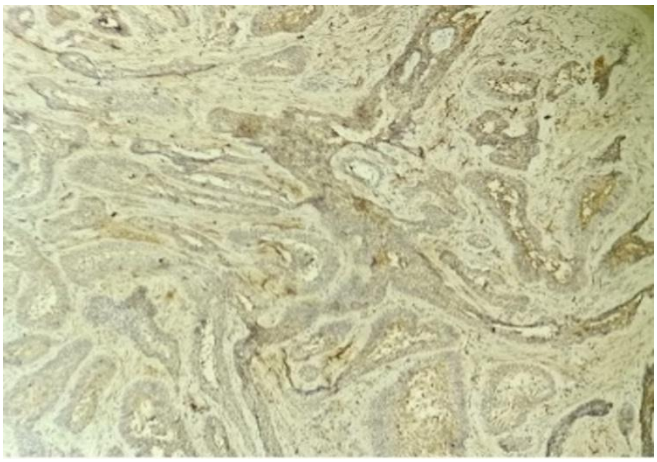

Fig. 4. CD 138 membrane-positive in tumor cells.

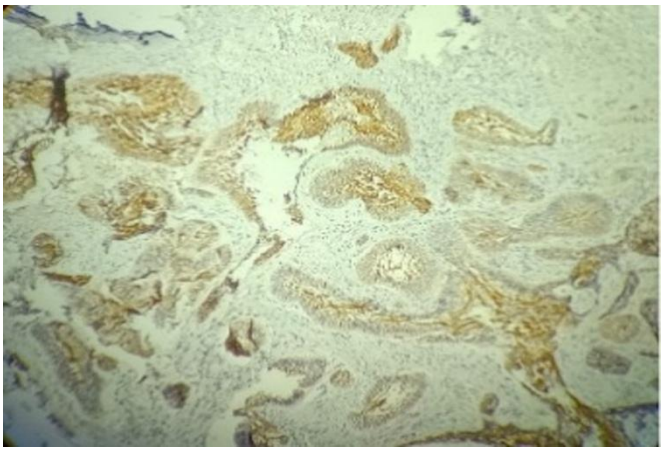

Fig. 5. CK 19 positivity in tumor cells.

3. Discussion

Ameloblastoma is a non-malignant tumor that grows aggressively in the areas of the body involved in tooth formation. It accounts for approximately 1% of all tumors found in the jaw, and it most commonly affects individuals between the ages of 30 and 50.[5] Research suggests that about 80% of these tumors appear in the lower jaw, while the remaining 20% occur in the upper jaw.[2] In many cases, the tumor may not cause any noticeable symptoms and may present as a slowly growing, hard, painless lump. On X-rays, lesions in the lower jaw typically show as clearly defined regions of bone loss, often

with a well-defined edge that may have a scalloped shape. Lesions in the upper jaw, however, tend to have less distinct borders. The internal structure of the tumor can vary, ranging from obvious areas to regions that include bony partitions.[9, 10] When examined under a microscope, Ameloblastoma is categorized based on different patterns, including follicular, plexiform, acanthomatous, basaloid, granular cell, and desmoplastic types. More than one type can be present in a single tumor.[11] The follicular type is the most common, accounting for approximately 32.5% of all cases, followed by the plexiform type (28.2%), the acanthomatous type (12%), and the desmoplastic type, which is less common, ranging from 4% to 13%. In the follicular type, the cells resemble those found in the inner layer of dental epithelium, characterized by tall, columnar cells with nuclei pushed away from the basement membrane. At the center of these structures, loosely arranged cells appear similar to the stellate reticulum. Interdigitating cords of epithelial cells characterize the plexiform type, which presents as an irregular mass. The acanthomatous type is linked to changes in the stellate reticulum, which transform into cells resembling squamous cells. The desmoplastic form is rare and characterized by a thick, fibrous layer surrounding compressed, angular clusters of tumor tissue.[5] The follicular type has the highest likelihood of returning after treatment, with a recurrence rate of 58.1%, while the plexiform type has a recurrence rate of 17.1%.[12] Immunohistochemical tests reveal that a protein called syndecan-1 (CD138), which is a type of sugar-coated protein, is commonly found in the stroma and stellate reticulum-like cells of Ameloblastoma. This protein helps cells adhere to each other, keeping them in place and preventing them from spreading. Additionally, other markers, known as CK19 and CK8, which are associated with epithelial cells involved in tooth development, are also present in all types of Ameloblastoma.

In our study, the ages of the patients were 47, 32, and 52 years, which aligns with findings from previous research.[5, 11] All three cases involved male patients. The first case was located in the anterior maxilla and presented as a multiloculated lytic lesion on radiographic imaging, with a follicular pattern observed under histological examination. Rajalakshmi et al. also reported a similar lesion with the same radiological features, exhibiting a mixed histopathological pattern that included follicular, acanthomatous, and desmoplastic components.[13] The second case was found in the posterior mandible and showed a cystic appearance on radiology. Histologically, it was identified as the desmoplastic variant of Ameloblastoma. Earlier studies have also reported similar findings, noting a mixture of histological subtypes.[14-16] The third case was located in the anterior mandible and also displayed a cystic, multiloculated lesion. Histologically, it showed a combination of follicular and plexiform variants of Ameloblastoma, which has been noted in other studies as well.[5, 17] All three cases were confirmed using immunohistochemical markers: CD138 and CK19.[11, 18] CD 138 demonstrated membranous positivity in both epithelial and stromal cells. CK 19, a marker specific to odontogenic epithelium, was positive in two of the cases. In the desmoplastic type, the odontogenic characteristics were reduced because the basal cells were negative for CK 19. Therefore, we observed that all three cases had different anatomical locations, distinct radiological presentations, and varied histological patterns.

4. Conclusion

Ameloblastoma is classified as a benign neoplasm; however, it is aggressive in nature and tends to recur. Diagnosis of Ameloblastoma is based on clinical, radiological, and pathological correlation. Very often, the diagnosis is made based on radiographic features; one should never rely on it alone. Histopathological features play a crucial role in the diagnosis and recurrence of Ameloblastoma. Early and accurate diagnosis can help oral surgeons take the necessary treatment options and prevent recurrence.

Conflict of Interest

The authors declared that there is no conflict of interest.

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