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Metastasizing Ameloblastoma

A 27-year-old man presented with an 8 cm diameter left maxillary mass and an enlarged cervical lymph node at levels II to III. There was a reported history of a previous unspecified operation on the maxillary mass which had yielded a diagnosis of ameloblastoma. Total maxillectomy with modified radical neck dissection was subsequently performed.

Microscopic examination of the maxillary mass shows epithelial islands and cords in a fibro-collagenous stroma. (*Figure 1*) The islands and cords are lined in the periphery by palisaded columnar cells with regular ovoid nuclei exhibiting reverse polarization. The nuclei are uniform with dispersed chromatin and no significant atypia. Towards the center of these islands are loosely arranged spindly to stellate cells (“stellate reticulum”). (*Figure 2*) Microscopic examination of the largest submitted lymph node shows an epithelial neoplasm with identical histologic features as the maxillary mass and a residual rim of lymphoid tissue at the periphery enclosed by the nodal capsule. (*Figure 3*) Similarly, there is neither atypia nor pleomorphism and only a few typical mitotic figures are seen in the nodal tumor. (*Figure 4*) These features support the diagnosis of a metastasizing ameloblastoma (MA).

Metastasizing ameloblastoma is rare with only about 70 reported cases.¹⁻⁵ Both the gnathic primary tumor and the metastatic foci have typical morphologies of a benign ameloblastoma with bland nuclei and absent to rare mitosis.^{6,7} There are no morphologic criteria that can predict this metastatic behavior. Thus, this diagnosis can only be made in retrospect upon the appearance of metastasis in a gnathic tumor that would otherwise have been diagnosed as a usual benign

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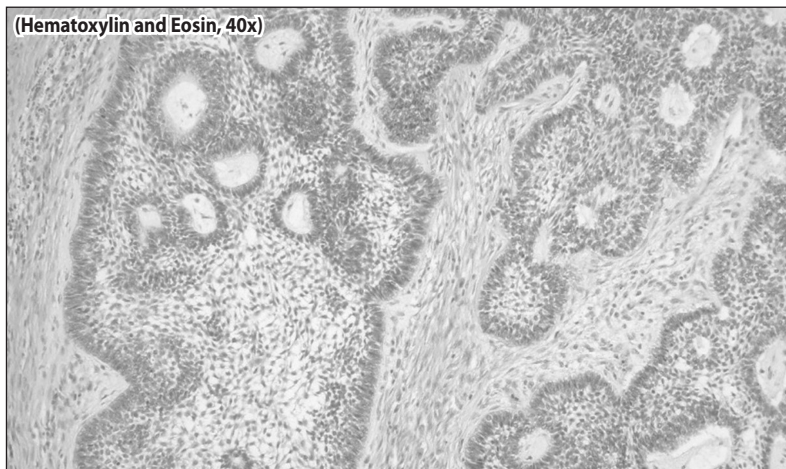


Figure 1. Section from the maxillary mass showing islands and cords of epithelial cells set in a fibro-collagenous stroma (Hematoxylin and Eosin, 40x magnification)

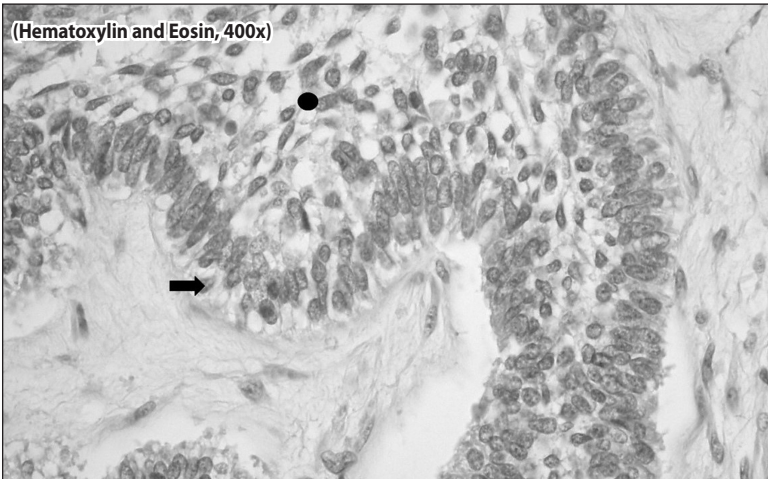


Figure 2. Peripheral palisaded columnar cells with reversed nuclear polarization (arrow) and central stellate reticulum (circle). The nuclei do not exhibit pleomorphism. (Hematoxylin and Eosin, 400x magnification)

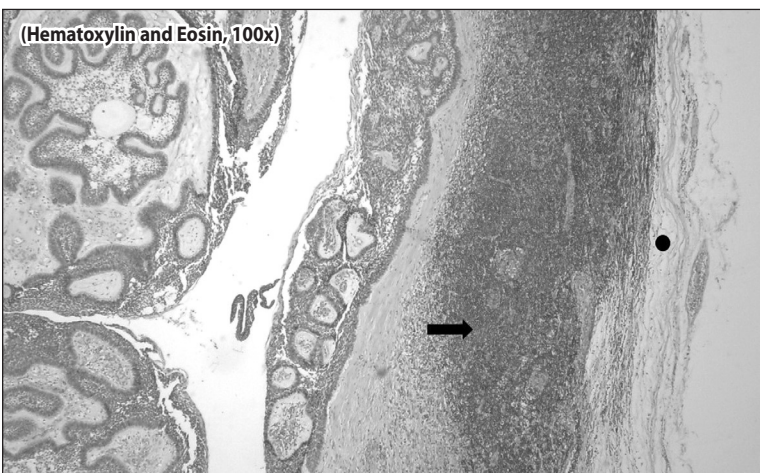


Figure 3. Section from the cervical lymph node containing metastatic tumor. There is residual nodal lymphoid tissue (arrow). The lymph node capsule is also shown (circle). (Hematoxylin and Eosin, 100x magnification)

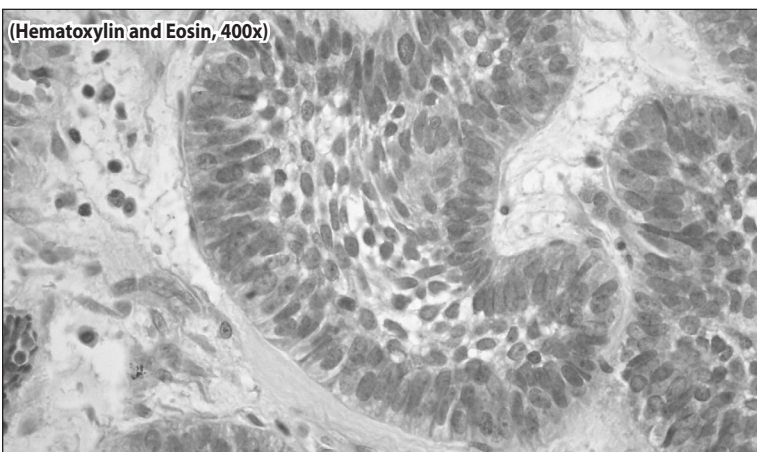


Figure 4. The tumor islands in the lymph node show a similar bland cytology. (Hematoxylin and Eosin, 400x magnification)

ameloblastoma.¹⁻⁷ Suggested risk factors include rapid tumor growth, large primary tumor, delay in treatment, mandibular site, prior radiotherapy and chemotherapy.^{4,5} However, what is most consistent in the literature is the history of multiple recurrences and multiple surgical interventions.^{1,4,5}

The differential diagnosis is an ameloblastic carcinoma (AC). Both MA and AC are considered as the malignant counterparts of ameloblastoma.⁷ However AC is characterized by the presence of the usual cytologic features of a malignant neoplasm such as nuclear pleomorphism, hyperchromasia and brisk mitotic activity – features that are lacking in MA.^{6,7}

The literature on metastasizing ameloblastoma lists the lungs (70 to 88% of cases) and the cervical lymph nodes (15-37.8% of cases) as the most common sites of metastasis.¹⁻⁵ Increased Ki-67 labeling and CD10 immunoreactivity have been reported to have significant correlation with recurrence.^{8,9} Whether these observations also apply to the risk of metastasis is not known due to the rarity of cases and hence the small subject populations of these studies.

As the risk of metastasis cannot be predicted by morphology, long-term follow-up appears prudent in all cases of ameloblastoma especially if characterized by recurrences and prior surgical interventions.

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