Metastasizing Ameloblastoma: A case report

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Abstract

Ameloblastoma is the most common odontogenic tumor, but the incidence of its metastasis is low. We report a case of recurrent ameloblastoma metastasizing to the cervical lymph node. This patient presented with a radiolucent cystic lesion in the left mandibular region. Segmental mandibulectomy along with cervical lymph node dissection was performed. Histopathological features revealed an ameloblastoma with single lymph node metastasis. The patient had two previous instances of lesion at same site which were diagnosed to be ameloblastoma on histopathology. Metastasizing ameloblastomas are different from ameloblastic carcinomas and lymph node metastasis is especially rare. The case is presented as it exhibits this peculiar feature.
Introduction
Ameloblastoma is the most common odontogenic tumor. Ameloblastoma has ability to develop metastasis in regional lymph nodes and distant organs. The lung is the most frequent metastatic site (88%) followed by regional lymph nodes (27%).[1]

In the WHO classification, a distinction is made between ameloblastoma, metastasizing (malignant), ameloblastoma and ameloblastic carcinoma. Malignant ameloblastoma differs from ameloblastoma due to the presence of metastases. They both have the same benign histology. However ameloblastic carcinoma has malignant cytologic features regardless of the presence of metastases.[2]

Case Report
A 38-year-old female, who 3 years prior had undergone successful surgical treatment of a left mandibular ameloblastoma, presented with a left submandibular swelling. Radiology confirmed a heterogenous solid-cystic nodule close to the previous lesion resection site. Left segmental mandibulectomy along with radical neck dissection was performed. A lytic solid-cystic lesion measuring 4 x 3 cm was seen. Five lymph nodes were dissected, largest of which measured 1.5 cm in diameter. Histopathology revealed a tumor with features typical of ameloblastoma. These included tumor cell islands with central stellate reticulum (Fig 1) and nuclei showing peripheral palisading with reversed nuclear polarity (Fig 2), anastomosing strands of cells, and a dense fibrotic stroma. A single lymph node containing metastatic deposits of tumor was noted (Fig 3). Metastatic deposits showed similar histomorphological features as that of the primary tumor.

Fig 1-Histopathology of Ameloblastoma- Primary tumor (tumor cell islands with central stellate reticulum)
Fig 2-Histopathology of Ameloblastoma- Primary tumor (nuclei showing peripheral palisading with reversed nuclear polarity)
Fig 3-Metastatic deposits of Ameloblastoma in cervical lymph node
Discussion
Metastasizing ameloblastoma is an ameloblastoma that metastasizes in spite of a benign histologic appearance. Its ICD-O code is 9310/3[3] In contrast, ameloblastic carcinoma are the tumors that show cytological features of malignancy like nuclear hyperchromasia and pleomorphism.

Metastasizing ameloblastoma shows no specific features different from ameloblastomas that do not metastasize. Therefore, this diagnosis can only be made in retrospect, after the occurrence of metastatic deposits. Thus, it is clinical behaviour and not histology that justifies a diagnosis of metastasizing ameloblastoma.[3] Despite its benign histological appearance, ameloblastoma has the ability to metastasize. Most metastatic deposits are seen in the lung with few reported cases of lymph node metastases.[4]

Due to the rarity of cases, treatment of ameloblastoma with metastases to lymph nodes is controversial. Some recommend radical neck dissection, while others have proposed simple excision with close follow up incases with a single node metastasis.[4]

Conclusion
Metastasizing ameloblastoma shows no specific features different from ameloblastomas that do not metastasize. Therefore, this diagnosis can only be made in retrospect, after the occurrence of metastatic deposits. Thus, it is clinical behaviour and not histology that justifies a diagnosis of metastasizing ameloblastoma. Metastasis is more likely to occur in recurrent tumors.

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References