Primary Ameloblastic Carcinoma of the Neck

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Ameloblastoma is an odontogenic usually benign tumor that rarely metastasizes to distant organs. Malignant ameloblastoma is defined as a histologically benign-appearing ameloblastoma with metastasis. Metastasis most commonly occurs in the lungs and lymph nodes. The case of a 54-year-old male is described, who presented with ameloblastic carcinoma on level I and II neck region. Clinical and radiological images confirmed no ameloblastoma in the jaw and no other primary tumor. We suppose that this case was metastatic ameloblastoma with unknown primary origin or ectopic ameloblastic carcinoma.

Ameloblastomas represent 1% of all tumors of the mandible and maxilla but occur more frequently in the mandible than in the maxilla (4:1 ratio).1 The ameloblastic carcinoma histologically demonstrates features of malignancy, such as pleomorphism, atypical mitoses, and a reverse nuclear–cytoplasmic ratio.2 The ameloblastic carcinoma behaves like a primary malignancy, with the ability for invasive local destruction, nerve involvement, lymph node spread, and distant metastasis.3-6 In this present case, a physical examination showed a firm, mobile, tender 4- to 5-cm-sized mass on the right submandibular area. A computed tomography scan showed a 4 to 6-cm-sized homogenous mass at level I and II of the right upper neck. The mass was compressing the submandibular gland anteriorly but no invasion to surrounding structures was detected (Figure 1). Positron emission tomography–computed tomography images showed a 5-cm-sized solitary mass at level I and II of the right neck with intense Fluorodeoxyglucose (FDG) uptake. No FDG uptake was observed by other head and neck regions, the lungs, or the mediastinum. We removed all level I to V lymphatic tissues, including the jugular vein and spinal accessory nerve. The low-power microscopic view revealed branching and anastomosing or papillary epithelial strands (Figure 2). The high-power view showed necrosis foci. More than 5 mitotic figures were counted per high-power field. The typical cytological features of ameloblastic differentiation include peripheral rim columnar cells that have nuclei displaced away from the basement membrane and contain small cytoplasmic vacuoles (Figure 2). The tumor cells were immunoreactive to cytokeratin (cytokeratin, ×100). Cytokeratin expression was accentuated on the peripheral rim because of hypercellularity. The tumor cells were also immunoreactive to vimentin (vimentin, ×100) and stained mainly in the peripheral rim. Postoperative radiation therapy was scheduled and the patient has remained disease-free.

In conclusion, the present patient revealed only a neck mass with ameloblastic carcinoma, and no primary lesion was detected in the head and neck region or the rest of the body, and his mandible and maxilla were completely normal. No previous study has reported on metastatic malignant ameloblastoma without a primary lesion. So, we suspect that this malignant ameloblastoma may have been a metastatic lesion with unknown primary origin or it originated from ectopic

Figure 1. Sagittal CT image (A) showing 5-cm-sized soft tissue mass on level I and II of the right neck and sagittal PET-CT image (B) showing neck mass with intense FDG uptake. Preoperative neck mass (C) showing protruding neck mass on the right upper neck and intraoperative finding after radical neck dissection (D) CT indicates computed tomography; FDG, Fluorodeoxyglucose; PET-CT, positron emission tomography–computed tomography.

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ameloblastic tissue in the neck. It is important to differentiate a metastatic derivation from a missed primary tumor or arising in ectopic ameloblastic tissue.

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