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Metastatic Ameloblastoma to the Liver: Rare Presentation of a Rare Disease

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Abstract

Ameloblastoma is a slow growing odontogenic epithelial neoplasm which originates from remnants of the dental lamina with a high recurrence rate, but a low tendency to metastasize. Locally invasive ameloblastoma is often aggressive and destructive, which erodes bone and invades adjacent structures. Despite a benign histology metastatic disease may occur and samples taken from metastatic tumor usually maintains the features of the original tumor. Ameloblastic carcinoma differs from ameloblastoma with malignant cytological features. Here we report an unusual case of ameloblastoma metastatic to lung and liver, unresponsive to systemic treatment with cisplatin and adriamycin, and well controlled with local surgical treatment.

Keywords: Ameloblastoma; Metastasis; Liver; Resection

Introduction

Ameloblastoma is a rare, benign or cancerous tumor of odontogenic epithelium. Locally invasive ameloblastoma is often aggressive and destructive, which erodes bone and invades adjacent structures. While these tumors are rarely malignant and progress slowly. A 23-year-old man is presented in this case report.

Case Report

A 23-year-old man presented with three months history of increasing lower abdominal discomfort and a change in bowel habits. According to the hospital records, he was a nonsmoker and denied alcohol consumption. He had had dental problems in 2004, underwent dental extraction and operations then. Following these procedures, he had noticed a painless swelling in his right mandible. The lesion was resected, and histopathology was reported ameloblastomatous. Between 2004 and 2015, the patient suffered 7 local relapses which were treated surgically. In 2015the patient started to have the above mentioned complaints. CT scans of the chest and abdomen revealed two metastatic lesions in the rightlungand ahuge liver masslocated in segments 2 and 3 (Figures 1 and 2). A core biopsy taken from this lesion was reported as metastatic ameloblastoma. Immunohistochemistry revealed diffuse positive staining with cytokeratin (CK) 14, CK 19, CK 5, beta-catenin, focal staining with CK 18 and negative staining with calretinin. The patient received six cycles of cisplatin and adriamycin doublet. CT scans taken after the 3rd and 6th cycles showed stable disease. Despite these three additional cycles of this combination was given, without achieving an objective response. The patient was discussed in our multidisciplinary

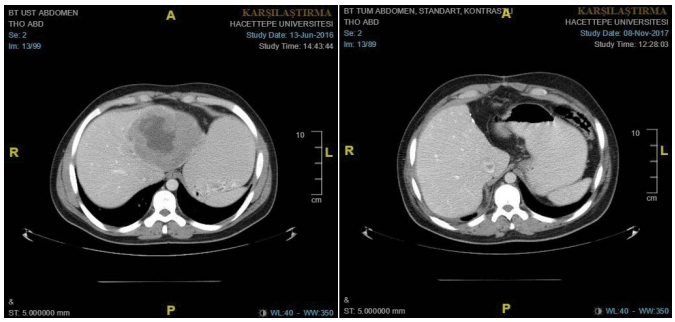

Figure 1: Preoperative and after resection of the liver lesion abdominal CT scans.

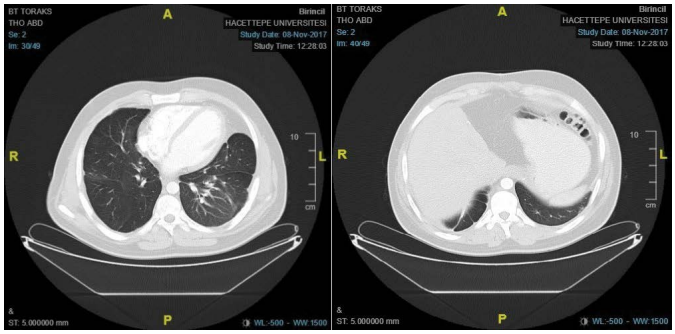

Figure 2: The patient thorax CT after resection of the pulmonary lesions.

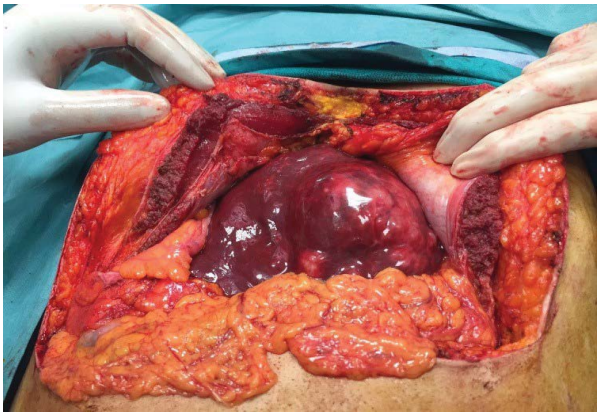

Figure 3: Intraoperative view of the metastatic lesion in the liver.

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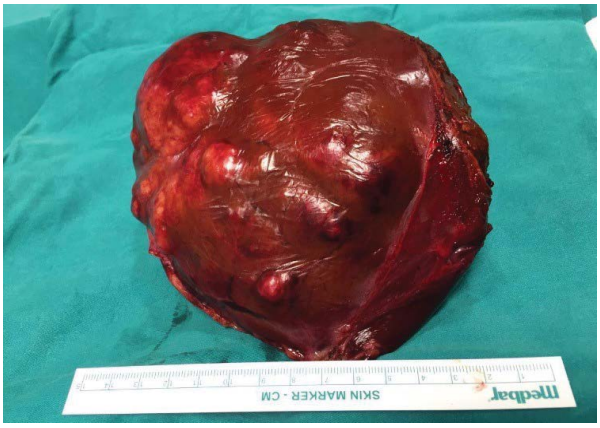

Figure 4: Metastasectomy specimen.

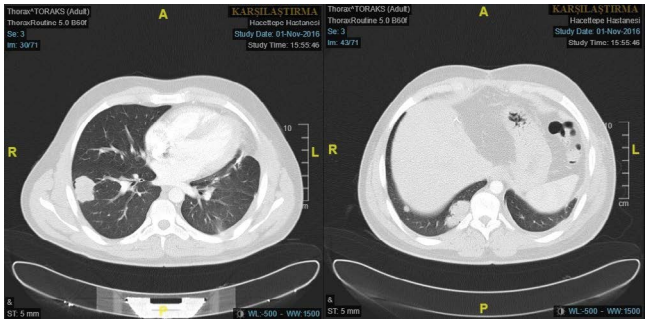

Figure 5: The patient thorax CT that shows metastatic pulmonary lesions (red arrows) pre-op.

tumor board and considering theresistance to systemic treatment, his good performance status, and reported success of local surgical approaches forthe disease, we decided to proceed with surgical treatment. In August 2016, the lesion in the liver was removed (Figures 3 and 4) and in September 2016, he underwent a right thoracotomy with resection of lung lesions (Figure 5). The pathological evaluation of all samples were consistent with ameloblastoma. Postoperative recovery was uneventful, and the patient is currently free of disease, at 15 months.

Discussion and Conclusion

Ameloblastoma is a rare, benign but locally aggressive odontogenic tumor which originates from the dental lamina and the odontogenic epithelium. The disease represents approximately 1% of all tumors of the maxilla and mandible. About 80% of the cases occur in the mandible [1]. The treatment of ameloblastoma is primarily surgery and the overall local recurrence rate of the disease with current treatment methods is approximately 10% [2]. Metastatic ameloblastoma is an infrequent entity, accounting for approximately 2% of ameloblastoma cases [3]. Ameloblastic carcinoma is a rare variant that may give rise to metastatic disease. The presence of locally extensive disease, duration of the initial tumor, multiple local recurrences and the type of the surgical procedures, exposure to radiotherapy and/or chemotherapy may all influence the development of metastatic disease [4]. Ameloblastoma may spread to distant sites and organs via the lymphatic, hematogenous routes and passively by aspiration [5,6]. Pulmonary spreading through hematogenous dissemination is the most commonly accepted route rather than aspiration. But, the presence of tumor in the bronchi and bronchioles does support the role of aspiration [7]. The most common sites of metastases in ameloblastoma are the lung, cervical lymph nodes, brain, and bone [8]. The lungs, as the most frequent site of metastasis, have been reported especially in patients undergoingmultiple operations (80%). Intraoperative tumor implantation via endotracheal tube is probably the cause in these cases. The patient being reported had a primary surgery and 7 surgical procedures for local recurrences, and therefore aspiration of tumor cells as a mechanism for metastasis cannot be ruled out. On the other hand, multiple pulmonary and liver metastases support to the theory of hematogenous dissemination. There usually is a long period from initial diagnosis to metastatic disease, somewhere in between 9 to 14 years, in various reports [5-10]. Metastatic disease in this case was detected 11 years after the initial diagnosis. Chemotherapy given at that time point was not effective and surgical resection was performed. The patient is free of disease at 15 months.

Ameloblastoma is a rare disease and establishing a standardtreatment approach will probably be not easy. Distant spread of the tumor is a challenging situation and surgery, if possible, is often the treatment of choice, with the most promising results [9,11]. Liver is an unusual metastatic site for ameloblastoma. The liver remains an exceptional and unusual metastatic localization of ameloblastoma, our case is, to our knowledge the second reporting this location.

Conflicts of Interest

The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication.

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