Postradiation Malignant Fibrous Histiocytoma of The Maxillary Sinus

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Malignant fibrous histiocytoma is the most common soft tissue sarcoma in adults. The common sites of involvement are the retroperitoneal space and limbs. The occurrence of this tumor in the head and neck is rare. Recent studies have shown that the occurrence of malignant fibrous histiocytoma in the head and neck can follow radiotherapy for the treatment of other tumors. We, herein, report a case of malignant fibrous histiocytoma, which developed 17 years after radiotherapy of a carcinoma in the maxillary sinus.

Keywords: Histiocytoma • malignant fibrous histiocytoma • maxillary sinus • radiotherapy

Introduction

Fibrous histiocytoma is composed of differentiated fibroblasts and histiocytes. A small percentage of these tumors behaves as a malignant lesion and is therefore called “malignant fibrous histiocytoma” (MFH).1, 2

MFH is the most common soft tissue sarcoma in adults. This term was first introduced in 1963 when it was thought this tumor originates from histiocytes.3 The results of new studies, however, indicated that the cell lines were immunophenotypically similar to primitive mesenchymal cells and currently its origin is considered to be from the fibroblasts.4, 5 The most common sites of involvement are the retroperitoneal space and limbs. The occurrence of this tumor in the head and neck is rare, and considering the site of involvement, it can cause pain, facial nerve palsy, hemoptysis, nasal congestion, and epistaxis.6 A case of maxillary sinus MFH was reported in whom this lesion at the beginning presented as a toothache.7 Recent studies show that the occurrence of MFH in head and neck, following radiotherapy, is more common.8 We, herein, report a case of MFH, which developed 17 years after radiotherapy of a carcinoma in the maxillary sinus.

Case Report

A 42-year-old man was referred to a private dental clinic complaining of pain in his upper left 6th molar area. On physical examination, an exophytic mass on the upper left bucco-gingival mucosa was found. The radiographic evaluation revealed a radiolucent area with ill-defined margins in the apical area of the left upper teeth extending to the maxillary sinus (Figure 1). The patient gave history of a lesion in the left maxillary

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sinus region 17 years before. Reviewing his medical chart revealed that the lesion had been an infiltrating anaplastic carcinoma (Figure 2a), which was treated by surgery and radiotherapy; the patient was then lost to follow-up.

Considering the history and present clinical findings, a magnetic resonance imaging (MRI) was advised. MRI showed a heterogeneous mass in the left maxillary sinus invading its anterior wall (Figure 3).

An incisional biopsy showed a low-grade spindle cell sarcoma, compatible with the diagnosis of fibrosarcoma. Giant cells with multiple nuclei were also the additional histopathologic features. After total resection of the mass, which measured 10×8×3 cm, the surgical margins were reported free of pathology. Immunohistochemical staining for vimentin, cytokeratin (CK), and EMA markers were done. Considering the negative results for the CK and EMA markers, and positivity for vimentin, the epithelial nature of the lesion was ruled out and the mesenchymal origin was verified.

On reviewing different histopathologic sections of the lesion, the diagnosis of MFH was considered. A whole body scan using $^{99}$Tc MDP and chest X-ray disclosed any evidence of lung metastasis. Four months later, the patient complained of the presence of an enlarged palpable right cervical lymph node. Fine-needle aspiration showed the presence of malignant cells. Axial and coronal computed tomography (CT) scans of the floor of the left orbit and left zygoma showed involvement of these areas, indicating recurrence of the lesion.

On maxillectomy, a 5×4×3 cm mass along the floor of the left orbit and nasal septum, were removed. On microscopic examination, atypical oval cells with clear hyperchromic nuclei were seen in a collagen matrix. There were numerous giant cells around the malignant cells. The number of mitotic cells was reported to be less than 10/HPF. The diagnosis of fibrohistiocytic neoplasm compatible with MFH (giant cell type) was finally considered for this patient (Figure 2b). After four months, on follow-up examination, metastases to the lung were noticed, and unfortunately, the patient passed away two months later.

Discussion

Sarcomas of the oral and maxillofacial region are rare. In a review of 32 cases of oral and maxillofacial sarcomas, reported over a 25-year period, only seven cases were attributed to MFH. In a review of literature for a period of 30 years, only four cases of head and neck MFH were
Approximately 1 – 3% of these tumors affect the head and neck, with about 100 cases reported to date.\textsuperscript{11} MFH of the jaws has a distinct clinical behavior, being very aggressive with a high rate of local recurrences and frequent metastases. The behavior of MFH is more aggressive than that of fibrosarcoma and osteosarcoma in this area.\textsuperscript{2} It occurs between the 5\textsuperscript{th} and 7\textsuperscript{th} decades of life; both sexes are equally affected by this tumor. The prognosis is worse for tumors occurring in the oral and maxillofacial region.\textsuperscript{12}

The results of a study in 1994 revealed that some patients developed MFH following radiotherapy for the treatment of head and neck tumors. This study showed that MFH would occur 2½ to 11 years after initial radiotherapy for treatment of soft or hard tissue tumors of the oral and maxillofacial region.\textsuperscript{3} Another report showed that radiation-induced sarcoma of the head and neck (RISHN) is a long-term complication of therapy. In 10 patients with features of RISHN, MFH was the most common pathologic diagnosis. The period of latency between initial radiation therapy and diagnosis of RISHN ranged from 9 to 45 years with a median of 17 years.\textsuperscript{13} The results of another study on radiation-induced MFH after radiotherapy of nasopharyngeal carcinoma revealed that the maxillary sinus is the most common site of occurrence of RIMFH.\textsuperscript{14} Considering the fact that our patient had a carcinoma of the left maxillary sinus around 17 years before, for which he had received radiotherapy, it was assumed that the cause of MFH in our patient could be attributed to radiotherapy.

Recent studies have shown that MFH occurring after radiotherapy, appears clinically as a red, lobulated, and tender mass which has a radiographically radiolucent appearance with ill-defined margins without periosteal or endosteal reactions.\textsuperscript{8} In a research on radiographic features of MFH, it was reported that the lesion has a relatively smooth surface and uniform density on intra-oral X-ray, and also CT images showing the clear separation of the tumor from surrounding soft tissues.\textsuperscript{15} This patient had similar clinical and radiographic features, but MRI revealed a heterogeneous mass in the left maxillary sinus invading the anterior wall of the sinus.

The presence of giant cells with multiple nuclei created confusion in microscopic diagnosis of MFH (giant cell type) and giant cell tumor (GCT). In this respect, others have also discussed the difficulties in the differential diagnosis of GCT and giant cell subtype of MFH. At the same time, it has been emphasized that MFH is a high-grade sarcoma displaying considerable more cellular atypia than GCT.\textsuperscript{16}

Unfortunately, 18 months after the diagnosis of MFH, our patient died following lung metastasis — a feature similar to that of other reports.\textsuperscript{17, 14} Involvement of the regional lymph nodes is reported in only 12% of patients.\textsuperscript{2, 14} In our patient, the initial signs of recurrence of tumor were also noticed in the right cervical lymph nodes.

It appears that radiotherapy for the treatment of tumors of the head and neck region can be considered as a risk factor for the development of malignant changes towards MFH.\textsuperscript{14, 18}

It is therefore recommended that patients with a history of receiving radiotherapy for treatment of the head and neck tumors, be followed periodically for detection of MFH or other RISHN.

**References**

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