Case Report

Recurrent malignant fibrous histiocytoma of vocal fold

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ABSTRACT

Malignant fibrous histiocytoma (MFH), which particularly involves the trunk and the upper and lower limbs, originates from the mesenchymal tissue. The larynx is one of the very rare locations. The disease has a poor prognosis due to local recurrence and metastasis. The first-line treatment is wide excision of the tumor. Herein, we report a case of MFH of the vocal fold in a 68-year-old man presenting with recurrence.

Keywords: Malignant fibrous histiocytoma, undifferentiated pleomorphic sarcoma; vocal fold.

Malignant fibrous histiocytoma (MFH), also known as a pleomorphic undifferentiated sarcoma, is the most common malignant soft tissue tumor in adults.[1] Its incidence is highest in the extremities and retroperitoneal region and it originates from the mesenchymal tissue.[1] The head and neck region are one of the rare locations in 3% of cases, and vocal fold MFH is an extremely rare neoplasm.[2-4] Vocal fold MFH often presents as a subepithelial nodule in the larynx.[5] This tumor has a high rate of local recurrence and metastasis and has been reported in a few cases in the literature until now.[6]

In this article, we present a case of vocal fold MFH of the larynx and discuss its diagnosis and treatment in the light of literature data.

CASE REPORTS

A 68-year-old man was admitted to our hospital with dysphonia which started after an upper respiratory tract infection and persisted for five months. He underwent microlaryngoscopic surgery due to vocal fold MFH two months previously. The patient reported no relevant precipitating factors in his personal or family history. An endoscopic examination of the larynx revealed the presence of a 0.4×0.4-cm polyp-shaped mass on the right vocal fold in the immediate posterior area of the anterior commissure (Figure 1). The movement of the vocal fold was normal. On palpation of the neck, no mass or abnormality was detected. His previous diagnosis was MFH. Computed tomography (CT) revealed an irregularity in the

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anterior portion of the right vocal fold (Figure 2). The histopathological report of a biopsy material taken from the free edge of the lesion was compatible with MFH. No distant metastases were observed on positron emission tomography (PET)/CT.

The tumor was totally excised by performing an extended type 4 cordectomy with a diode laser. The microlaryngoscopic diode laser procedure was performed under general anesthesia by surgical microscopy with a 400-mm focus and direct laryngoscopy according to the Kleinsasser technique using a contact diode laser (30 W, 980 nm) (Biolitec Ceralas, CeramOptec, Bonn, Germany). The mode of laser operation was a continuous wave with variable power (~10 W) delivered to a flexible optical fiber (≥300 μm).

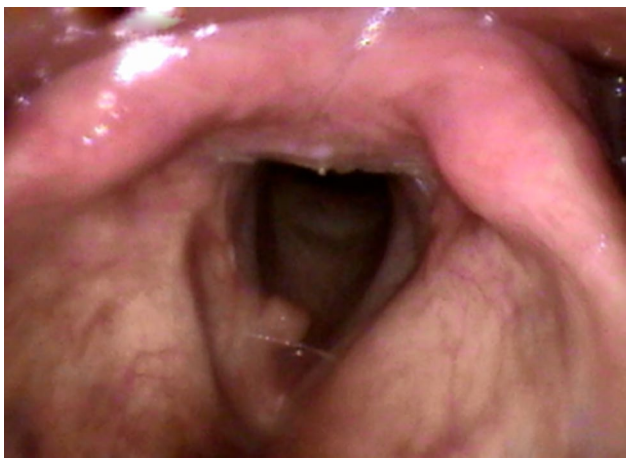

Figure 1. *Preoperative endoscopic view: the polypoid mass arising from the right vocal fold near the anterior commissure.*

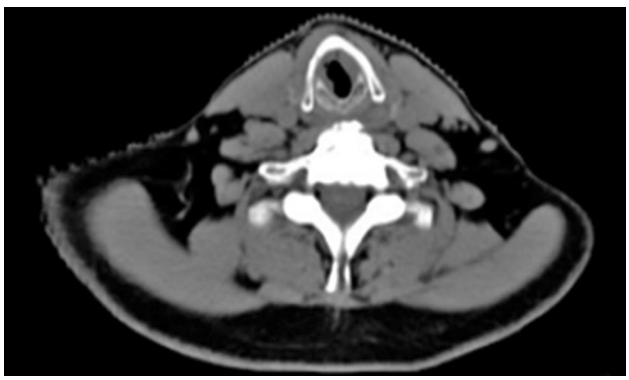

Figure 2. *Preoperative computed tomography view: Right vocal fold had irregularity (axial section)*

Surgery was completed after confirming the absence of tumors in the margin. No tracheotomy was performed. No early or late complications occurred.

The histopathological findings of the excised specimen were compatible with MFH. Postoperative pathological examination also confirmed that the surgical margins were negative. The histopathological examination using the hematoxylin-eosin staining showed that the surface of the MFH tumor was covered with a squamous epithelium which was partly eroded (Figure 3). Immunohistochemical examination revealed positively diffused vimentin staining in the spindle tumoral cells (Figure 4).

According to the decision of the Head and Neck Cancer Council, postoperative radiotherapy (RT) was planned, considering the high incidence of local recurrence in this patient. The patient was irradiated with a volumetric arc intensity modulated RT (IMRT) technique. A total of 33 fractions and three planned target volumes (PTVs) of RT were given (PTV 50, PTV 54, and PTV 66). A slight edema appeared after RT. A subsequent examination for metastasis revealed no abnormalities. Endoscopic examination revealed no evidence of recurrence during two-year postoperative follow-up. A written informed consent was obtained from the patient.

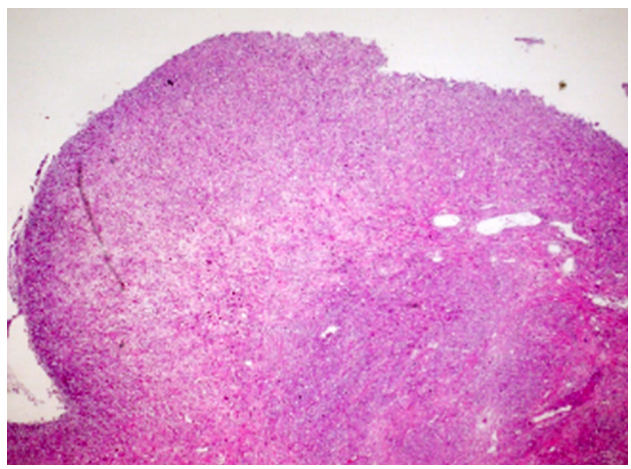

Figure 3. *Histopathology shows that the surface of the malignant fibrous histiocytoma is covered with squamous epithelium, which is partly eroded (H-E, ×40).*

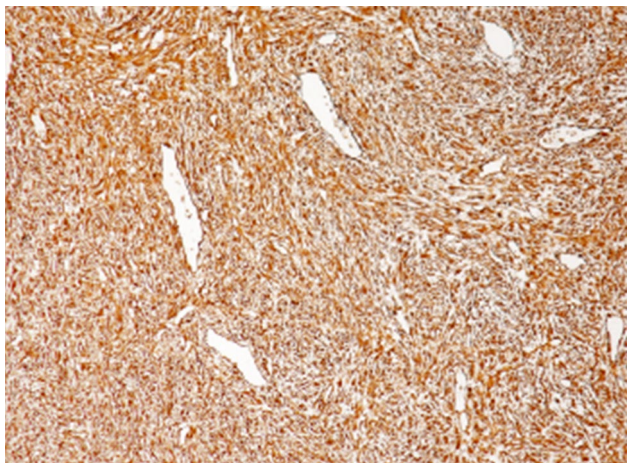

Figure 4. Findings on histopathology: Immunohistochemistry shows that the cells are positive for vimentin (×200).

DISCUSSION

Malignant fibrous histiocytoma is an aggressive tumor with a quite high recurrence rate of 44% reported in the literature.[7] The rates of metastasis to the lungs and lymph nodes are also high as 82% and 32%, respectively.[7,8] To date, very few cases of laryngeal MFH have been reported in the literature, and this tumor location accounts for only approximately 1% of all MFH cases.[2,3] Malignant fibrous histiocytoma usually affects patients between the ages of 50 and 70 years and is more common in men.[9] The major prognostic factors are the clinical stage, as defined by the tumor grade, size, and depth of involvement, and the presence of distant metastases.[7,8]

More common lesions are encountered frequently in the otolaryngology clinics and should be differentiated from vocal fold polyps. These lesions include laryngeal cysts, laryngeal papillomas, vocal fold hematomas, and early malignant tumors.[1] Differential diagnosis is difficult; therefore, these lesions are mostly diagnosed pathologically after local resection. Kim et al.[1] reported that the image of the MFH tumor resembled that of vocal fold polyps. In our case, surgical exploration revealed a solid, round mass which was firmly attached to the vocal fold.

Radiological findings are usually non-specific in cases of MFH of the head and neck region, and the tumor margins are not completely clear radiologically.[4] In our case, the value of imaging was limited, as the tumor was very small. The vocal fold had also irregularities and thickening.

Malignant fibrous histiocytoma neoplasm is the most common soft tissue sarcoma of late adult life. It can be categorized into five main subtypes: *(i)* storiform/pleomorphic, *(ii)* myxoid, *(iii)* giant cell, *(iv)* inflammatory, and *(v)* angiomatoid. The most common subtype is the storiform/pleomorphic subtype, which accounts for 50 to 60% of all these tumors.[10] Differential diagnoses include pleomorphic rhabdomyosarcoma, fibrosarcoma, spinocellular carcinoma, angiosarcoma, hemangiopericytoma, pleomorphic liposarcoma, and lymphoma.[11]

In addition, MFH tumors originate from the interstitial cells which differentiate fibroblasts and histiocytes.[12] Histologically, they exhibit a diverse cellular composition, as the fibrosarcomatous and histiocytomatous components of the tumor are present in a mixed fashion.[1] In our case, histological examination showed the storiform/pleomorphic subtype.

In general, surgery is the primary treatment option, and most recurrences of MFH are attributable to insufficient resection.[5,13] Frozen-section margins should be accurately confirmed histopathologically. Since very few cases are reported in the literature, it is difficult to draw any clear conclusion regarding the prognosis of vocal fold MFH. A wide surgical excision with a safe margin of at least 3 mm is the first treatment option in early stage MFH of the vocal fold.[1] Sabesan et al.[6] reported that marginal resection of MFH of the head and neck had a local recurrence rate of up to 85%, whereas radical resection had a local recurrence risk of only 27%. Therefore, resection with a wide surgical margin is recommended. Several reported cases of MFH of the larynx were treated by total laryngectomy.[2,3,14]

A combination of RT and chemotherapy does not have any further advantages compared to surgical treatment.[5] Although local RT and adjuvant chemotherapy can significantly increase survival rates and reduce the risk of metastasis in head and neck tumors,[8,14] RT in

MFH is used in patients with high risk, in non-operated patients with recurrence, and in cases of distant metastasis. If an incomplete resection is performed, postoperative RT should be considered.[11,14] In the present case, although the surgical margins were negative, the recurrence of the tumor led us to consider postoperative RT.

In conclusion, postoperative RT can be recommended in cases of MFH with recurrence. If a complete resection cannot be performed and metastases are present, the patient should be closely followed in the long-term.

Declaration of conflicting interests

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