A 54-year-old man who was a nonsmoker presented at our ear, nose, and throat (ENT) clinic complaining of progressive hoarseness for the preceding 3 months. Fiberscopy revealed a 2 mm × 2 mm right-false vocal fold nodular lesion (Figure 1A). Direct laryngoscopy with biopsy was performed, and a diagnosis of supraglottic squamous cell carcinoma (SCC; stage I) was made. The patient completed radiotherapy (66 Gy in 33 fractions) and was then regularly followed up at our ENT clinic.

During a routine follow-up 3 years after completion of radiotherapy, a left glottic lesion was found during fiberscopic examination. The patient was closely followed at our clinic over the subsequent 5 months, during which the lesion progressed (Figure 1B and C). Biopsy of the lesion was suggested at that time but the patient declined. Two months later, the patient was brought to our emergency department because of dyspnea and noisy breathing. Contrast-enhanced computed tomography of the larynx revealed a laryngeal space–occupying lesion that severely compromised his airway. Emergent tracheotomy was performed within a few hours, and tumor tissue was obtained through laryngoscopic biopsy. The pathology report suggested atypical spindle cell tumor with hyperactive mitosis and nuclear pleomorphism. Based on the clinical presentation, malignancy was highly suspected. After discussion with the patient and his family, total laryngectomy without neck dissection was performed followed by reconstruction with radial forearm free flap. Under gross inspection of the specimen, the left glottic tumor was approximately 3.5 cm in diameter and extended to the supraglottis and subglottis (Figure 2). Surgical pathology confirmed the diagnosis of malignant fibrous histiocytoma (MFH). The tumor was composed predominantly of aberrant spindle cells with abundant fibrous stroma (Figure 3A). Immunohistochemistry (IHC) evaluation showed strong expression of vimentin and smooth muscle actin but only localized weak expression of cytokeratin 5/6 and cytokeratin AE1/AE3 (Figure 3B and C). These findings supported the diagnosis of radiation-induced MFH. Because of a clear surgical margin and lack of high-risk features, we did not advise adjuvant therapy. The patient has been followed at our clinic and is currently free from recurrence (7 months since total laryngectomy).

Malignant fibrous histiocytoma is a type of soft tissue sarcoma comprised of poorly differentiated spindle-shaped mesenchymal cells. Malignant fibrous histiocytoma occurs most commonly in limbs or the retroperitoneum and is relatively uncommon in the head and neck region.1 Laryngeal MFH is rare, representing approximately 3% of all laryngeal malignancies.2 Because MFH lacks specific symptoms or features of gross appearance, IHC is crucial for distinguishing MFH from SCC. In cases of MFH, IHC staining is typically positive for mesenchymal markers (eg, vimentin and smooth muscle actin) and negative for epithelial markers (eg, cytokeratin 5/6 and cytokeratin AE1/AE3).3 Conversely, in cases of SCC, IHC staining yields opposing results.
Approximately 3% of all cases of MFH arise at the site of previous radiotherapy and are considered to be radiation-associated sarcoma (RAS).\(^1,2\) Compared to sporadic sarcoma, RAS has a far less favorable prognosis.\(^2\) The genetic etiology of RAS and sporadic sarcoma are presumed to be different; however, no reliable and widely used marker is available to distinguish these 2 entities from each other; differentiation between them depends mainly on clinical judgment. For every tumor found in a previously irradiated area, the possibility of RAS should be considered until proven otherwise. In the original version of the RAS diagnostic criteria established by Cahan et al in 1948, the latency period was defined as at least 5 years.\(^4\) Arlen et al suggested a shorter latency period (3 years), which has since been widely applied in the literature.\(^5\) In our case, the latency period between radiotherapy and subsequent RAS occurrence was approximately 3 years.

For nonmetastatic laryngeal MFH, the mainstay of treatment is complete surgical resection. Margin status is one of the most pertinent modifiable prognostic factors. Survival has been noted to be considerably better if RAS is resected with a microscopically clear margin.\(^6\) Total laryngectomy is often required to achieve complete resection. In some carefully selected cases, transoral laser microsurgery (TLM) might be sufficient for complete tumor excision.\(^7\) More attention should be given to patients who undergo TLM for laryngeal MFH because assessing their margin status is relatively difficult.

Adjuvant radiotherapy can be considered if a clear margin has not been achieved. However, in cases of RAS, radiation toxicity is expected to be more serious because of the cumulative dose from the previous exposure. A role for chemotherapy for RAS has not been established because few studies have demonstrated that it offers a benefit.

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