Malignant laryngeal fibrous histiocytoma: a new concept of management

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A 75-year-old male patient presented to the Speech and Language Disorder Outpatient Clinic of King Abdullah Medical City, Makkah, Saudi Arabia, on February, 2010, with a complaint of gradual onset and progressive course of dysphonia for 2 years. Nasofibrolaryngoscopic examination indicated a polipoidal mass of about 1.5 × 1 cm masking the entire glottic inlet. The mass was removed by direct laryngoscopy and diagnosed as a malignant fibrous histiocytoma. The patient developed recurrence of the tumor on the same site postoperatively and underwent the same procedure of excision, followed by a second recurrence. Direct laryngoscopic excision of the tumor was performed for a third time, but with a safety margin of 2 mm. Monthly follow-up examination indicated no postoperative recurrence till 10 months.

Keywords:
fibrous histiocytoma, laryngeal cancer, laryngoscopy, malignant

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Introduction

The term malignant fibrohistiocytoma (MFH) was suggested by Kauffman and Stout [1]. The incidence of MFH was reported to be less than 2% of all malignancies in the larynx and accounted for ~5% of all sarcomas of the head and neck [2]. It has a non epithelial origin, and around 3% of such tumors occur in the area of the head and neck, and is one of the rare neoplasms of the larynx [2,3]. Hardly 50 cases of laryngeal MFH have been published in the medical literature to date [3,4]. In laryngoscopy, the most common presentation is that of epithelial nodules or polyps [5].

MFH is a tumor that has a mesenchymal origin and uncertain histiogenesis. Histologically, there are five subtypes of MFH [6,7]. The histological diagnosis is made on the basis of the proliferation of neoplastic fusiform and polygonal cells that resemble fibroblasts and histiocytes with aberrant atypia and mitosis. These are organized in bundles with a variable pattern. It is difficult to establish a correct diagnosis with conventional histological techniques and to be sure, we need confirmation from immunohistochemistry (IHC) to distinguish it from other tumors with mesenchymal differentiation and spindle cell carcinoma [7–9]. In our case, IHC was very useful.

Case report

A 75-year-old male patient presented to the Speech and Language Disorder Outpatient Clinic of King Abdullah Medical City, Makkah, Saudi Arabia, in February 2010 with a complaint of gradual onset and progressive course of dysphonia for 2 years. On physical examination, no enlarged or affected lymph nodes were found in the neck and computed tomographic scan neck also showed the same finding, that is, no neck lymph nodes. Nasofibrolaryngoscopic examination and documentation indicated a huge polipoidal mass of about 1.5 × 1 cm masking the entire glottic inlet, which impeded the identification of its origin (Fig. 1a). The research was approved by institutional review board of King Abdullah Medical City and signed consent form got from the patient.

The mass was removed by direct laryngoscopy in the operation theater and sent to the Anatomical Pathology Unit. Histopathological examination indicated an infiltrating malignant neoplasm beneath the intact squamous surface epithelium. It was composed of pleomorphic polygonal to spindle cells showing hyperchromatic to vesicular nuclei with prominent nucleoli and moderate to abundant cytoplasm. These cells were arranged in the form of nests, fascicles, and vague whorls with a focal storiform pattern. Numerous gapping blood vessels were noted in fibrous stroma. There were many tumor giant cells with brisk mitotic activity and many abnormal mitotic figures. The IHC showed that the tumor cells were positive for vimentin and CD68, but negative for CK AEI/AE3, EMA, CK5/6, HMWCK, P63, HMB45, synaptophysin, CD57, desmin, muscle-specific action, S100, and CD34 (Fig. 2a–d).

A postoperative follow-up examination indicated the presence of a small recurrent mass at the same site as the previous mass on the anterior part of the left vocal fold (Fig. 1b).

We discussed with the patient and his family the possibility of further extensive surgical intervention, but they refused it as they feared that it would affect his life style adversely, considering his old age. Regular monthly follow-up examinations further indicated that the mass began to increase in size locally after 8 months in October 2010 (Fig. 1c).

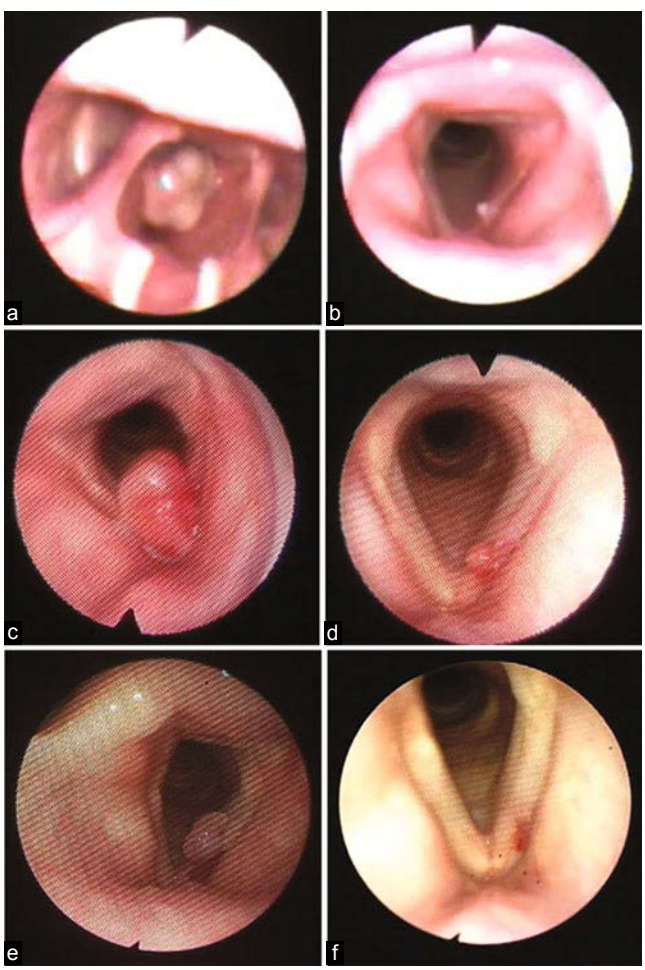
Afterwards, a direct laryngoscope excision was performed in March 2011 and histopathological diagnosis indicated the same diagnosis as MFH. The patient continued regular monthly follow-ups. After 4 months, a small recurrence appeared on the same place at the anterior one-third of the left vocal fold (Fig. 1d).

The lesion remained stable in size for 1 year afterwards and the size started to increase slowly up to 5 × 5 mm (Fig. 1e).

Direct laryngoscopic excision was performed again, but on this occasion, we removed the tumor with a safety margin of about 2 mm on January 2013. Monthly follow-up examination indicated no recurrence until January 2015 (Fig. 1f).

During the last follow-up examination, there were no enlarged or palpable lymph nodes in the neck.

Figure 1



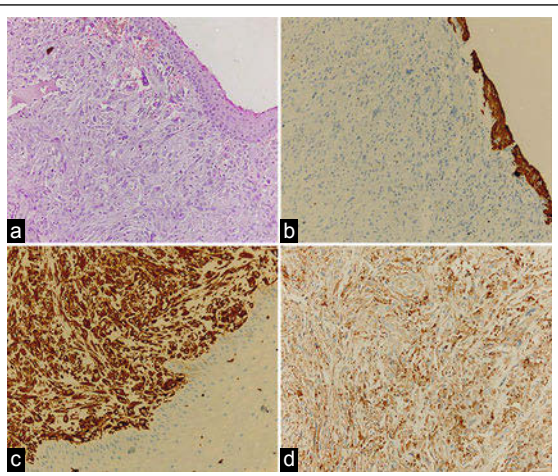
(a) On first examination. (b) Postoperative examination after the first surgery. (c) Preoperative examination before the second surgery. (d) Postoperative examination after the second surgery. (e) Preoperative examination before the last surgery. (f) 10th month postoperative examination after the last surgery.

Discussion

The first case of laryngeal MFH was described in 1972 [7]. No evidence-based guidelines for the management of these tumors were established because of their scarcity [10]. It was reported that laryngeal MFH is more common in men than in women, with a ratio of 3 : 1. It appears in adults with an average age of 43 years, but also has a very wide range from newborns to 93 years [6]. Two male cases were reported in 2004, 54 and 67 years of age, respectively [9]. Our case was a 75-year-old man. Dysphonia is the most common primary symptom; dysphagia and dyspnea can also be observed depending on the size and the localization of the lesion [4]. Our case presented with dysphonia and dyspnea as the lesion was very large, closing most of the glottic inlet, but the other two times, he was complaining only of mild dysphonia. However, hoarseness was the most common primary symptom in one case reported in one study [9].

Polipoidal lesions in our case were almost identical to two similar cases reported in 2004 by Bish *et al.* [9]. A globular mass without establishment of the lower limit was found by indirect laryngoscopic assessment in 2004. No neck nodes were clinically palpable. An

Figure 2



(a) Hematoxylin and eosin stain (×10): malignant fibrous histiocytoma showing highly atypical spindle cells arranged in fascicles and a few giant cells. (b) CK AE1/AE3 immunostain (×10): tumor cells are negative for CK AE1/AE3. (c) Vimentin immunostain (×20): tumor cells are strongly positive for vimentin. (d) CD68 immunostain (×10): tumor cells are moderately positive for CD68.

emergency tracheostomy was considered optimal, which resulted in marked improvement as the patient's distress worsened on lying supine [11]. This was similar to our case in its first presentation, but fortunately, transnasal intubation was performed successfully for completion of the procedure.

The histological differential diagnosis in this case was spindle cell carcinoma, pleomorphic sarcoma including MFH, neuroendocrine tumor, and malignant melanoma. The immunohistochemical studies showed negative staining for all epithelial markers (CK AEI/AE3, EMA, CK5/6, HMWCK), thus arguing against the diagnosis of spindle cell carcinoma. Negative staining for HMB45, synaptophysin, CD57, desmin, muscle-specific action, S100, and CD34 excluded the possibility of malignant melanoma, neuroendocrine tumors, and other specific types of pleomorphic sarcomas. The diagnosis of MFH was made after excluding all the other possibilities and positive staining for CD68 and vimentin. Therefore, a definitive diagnosis is difficult with conventional histological techniques and requires confirmation by IHC [9].

For MFH, simple radiotherapy or chemotherapy was not recommended, whereas comprehensive therapy combined with surgery was considered the best choice. Surgery is the first choice of treatment for the majority of authors. The technique used varies depending on the size and localization of the tumor, as with epidermoid carcinomas. Sabesan *et al.* [12], suggested that radical resection of a tumor is a more efficacious method for improving survival and reducing recurrence. However, partial laryngectomy and laryngomicrosurgery maintain laryngeal function and improve quality of life following surgery, and are recommended in early MFH [3,10]. Segmentary removal with wide resection margins is also advisable and in those cases where a conservative treatment is decided, it is mandatory to establish close follow-ups to detect possible recurrences as soon as possible [9].

Treatment methods of the neoplasms are radical surgery, radiotherapy, chemotherapy, and associated methods of therapy. Laser surgery (chordectomy) was used to treat this tumor without the need for radiotherapy [13].

Some authors suggest chemotherapy for other types of sarcomas [8]. The plan of treatment in our case was directed by the patient himself and his family, who agreed to a direct laryngoscopic excision. However, in the last instance of excision, where the safety margins were excised and the patient had no recurrence until, 10 months postoperatively. As per his choice, the patient's style of life was not adversely affected, except for three simple surgical daycare simple procedures; thus this must be considered as a management plan for MFH.

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This manuscript has been read and approved by all the authors, the requirements for authorship have been met, and each author believes that the manuscript represents honest work.

Conflicts of interest

There are no conflicts of interest.

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