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Case Report

Liposarcoma of the larynx: report of a case and review of literature

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Abstract: Liposarcomas of the larynx are extremely rare tumors, with only 37 cases reported in the English or French literature to date. The first two cases of laryngeal liposarcomas were reported respectively by Kapur and Dockerty in 1968 [1, 2]. Liposarcoma of the larynx is at high risk of local recurrence and seldom has metastatic potential. Prognosis for this tumor is better than that of non laryngeal liposarcoma. The present case is the first patient of primary liposarcoma of the larynx reported from China. A review of the literature was performed, and the presentation, position, pathological diagnosis, treatment and prognosis of the patients with liposarcoma of the larynx of the reported cases before are analyzed.

Keywords: Larynx, liposarcoma, surgery, radiotherapy, chemotherapy

Introduction

Liposarcoma is one of common soft tissue malignant tumor and is often found in the lower extremities and retro peritoneum. Only about 5.6% of liposarcomas are found in the head and neck, and most of the tumors arise from the soft tissues of the neck. Laryngeal liposarcoma (LLS) is extremely rare, with only 37 cases reported in the English or French language literature. In the present study, we report a case of LLS and analyze the 37 cases of LLS.

Case report

A 53-year-old man had a history of heavy smoking for 20 years and chronic laryngitis for 2 years, with airway obstruction that had developed over 3 months. A so-called laryngeal polyp had been removed from the right arytenoid region, aryepiglottic fold (AEF) and false vocal fold (FVF) 3 months before by surgical excision in a county hospital. According to the clinic doctor, the neoplasm was a yellow-grey 1×1 cm polypoid mass and was unencapsulated. The patient did not take the pathological examination in the country hospital. One week before, the tumor recurred. A laryngoscopic examination disclosed the irregular tumor mass in the right arytenoid region, AEF and FVF (**Figure 1**). We received irregular scattered masses excised from the focus with a size of 0.8×1 cm in large diameter.

The specimen was examined histologically and immunohistochemically. The tumor was composed of fat lobules with a few fibrous tissues. The adipocytes were of different size with scattered lipoblasts. At high-power view, well differentiated liposarcoma was characterized by adipocytes with a great variation in size and multivacuolated lipoblasts (**Figure 2A-C**). No mitotic figures in the areas of well-differentiated liposarcoma and no atypical spindled cells were seen. Immunohistochemistry on formalin-fixed paraffin-embedded tissue revealed positivity of the tumor cells for vimentin, S-100 and MDM2 protein (**Figure 2D-F**). Tumor cells were negative for AE1/AE3 cytokeratin, myoglobin, and CD68 immunostains. Final pathological diagno-

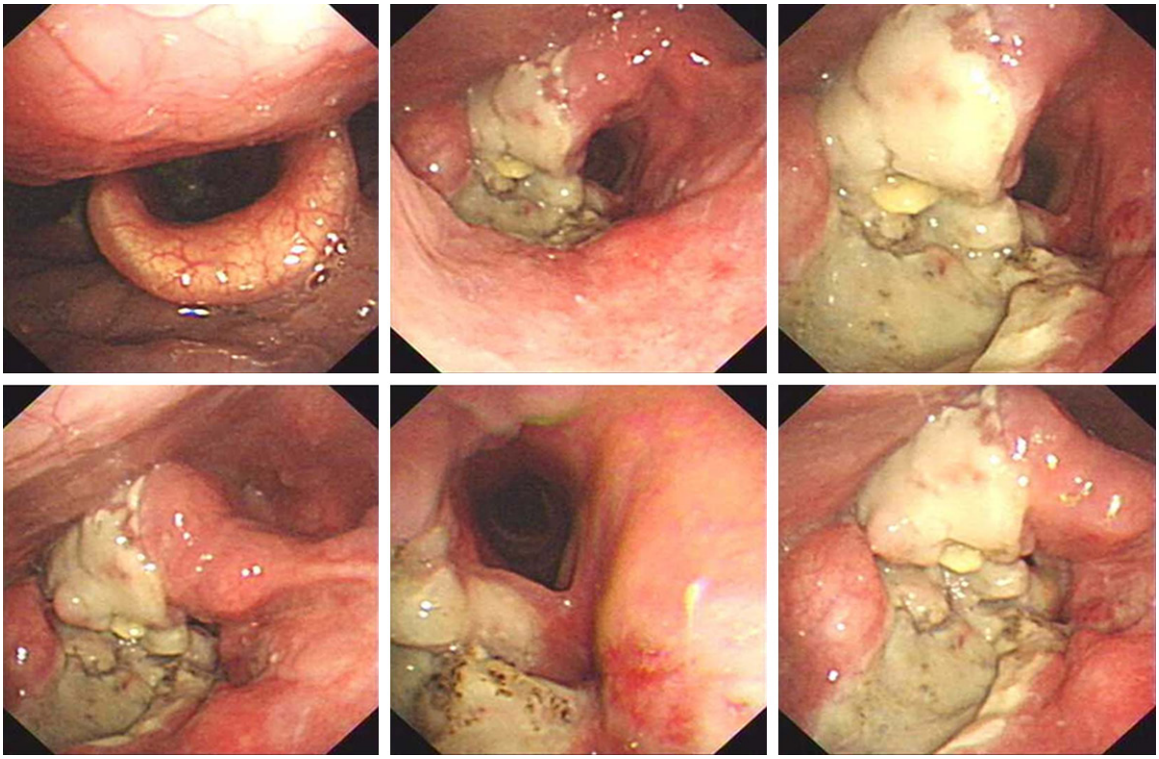

Figure 1. Laryngoscope showed the tumor mass in the right arytenoid region, aryepiglottic fold (AEF) and false vocal fold (FVF).

sis, suggested by the histological appearance and immunohistochemical profile, was atypical lipomatous tumor/well-defferentiated liposarcoma.

As a consequence, total laryngectomy was performed. The study of the latter specimen revealed tumor infiltration of the tumor margin. The patient did not receive radiotherapy and chemotherapy. Today, 13 months after laryngectomy, he is alive and well, without any evidence of recurrence or metastases.

Discussion

Liposarcoma, which is much less frequent than lipoma, was first described by Virchow in 1857 [26]. Laryngeal liposarcomas (LLS) are exceedingly rare, with only 37 cases been reported in the English or French language literature to date. Having reviewed the 37 cases of LLS reported (see **Table 1**), we summarize the clinical features are as follows: LLS are more common in men, and only 4 cases have been reported in women (male to female ratio is 8:1). The mean age of the patients is 55 years (ranged from 28 to 83 years) in the reported cases. Most of the reported cases affected the supraglottic area. There are only 4 cases affecting the true vocal cords. The most frequent complaints at presentation are airway obstruction, snoring or dysphagia, although some patients present because of hoarsenessor throat discomfort. Smoking has been suggested as an environmental factor in the development of this neoplasm.

The World Health Organization pathological classification (WHO 2013) identifies four histological subtypes of liposarcoma: well differentiated/atypical lipomatous tumor, myxoid/round cell, pleomorphic and dedifferentiated. We present a case of a laryngeal well differentiated liposarcoma. To our knowledge, this is the first case of laryngeal liposarcoma reported from China.

Histologically, most of the LLS are low grade tumors (either well-differentiated or myxoid liposarcomas), with only few reports describing high grade tumors (pleomorphic, myxoid/round-cell or dedifferentiated liposarcomas) [2, 5, 6, 17].

The main differential diagnosis of LLS is lipoma and myxoid chondrosarcoma. Immunohistoch-

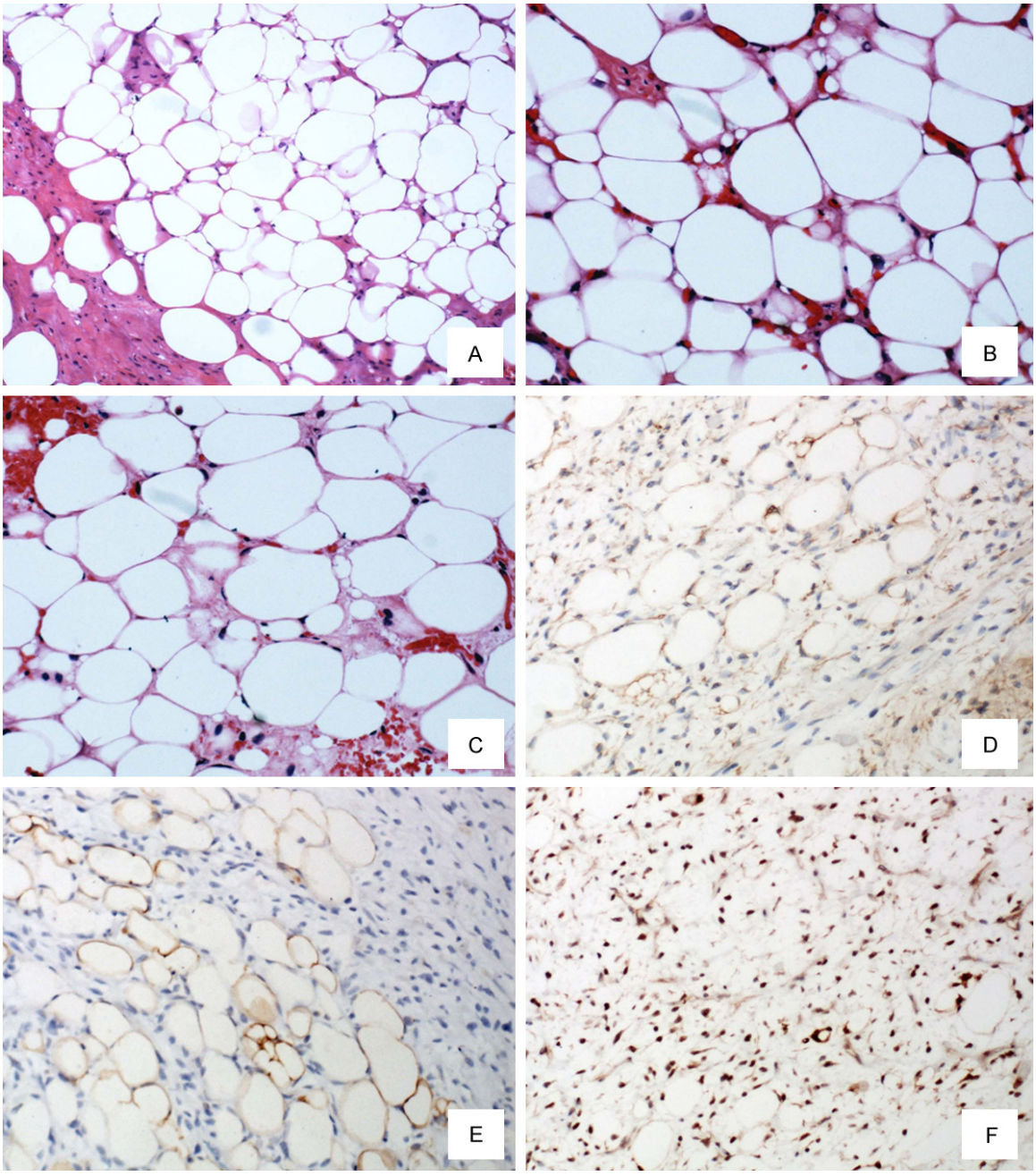

Figure 2. Microscopically, the tumor consisted of well-differentiated liposarcoma (A, hematoxylin-eosin, ×100). At high-power view, well-differentiated liposarcoma was characterized by multivacuolated lipoblasts (B and C, hematoxylin-eosin, ×200). On immunohistochamical analysis lipogenic areas showed positivity for vimentin (D, ×200), S100 (E, ×200) and MDM2 (F, ×200).

emistry cannot distinguish them, because both lipoma and chondrosarcoma are positive for S100 protein. Therefore, pathologists should find the typical histological features of liposarcoma (mainly the presence of lipoblasts) and make sure that there are no chondromatous areas. Lipomas are usually well-demarcated, encapsulated lesions which have no tendency to infiltrate into the surrounding normal tissues, show no lipoblasts and allow a simple excision without recurrences.

Most LLSs are indolent with low-grade histology. They are locally aggressive but have no tendency to spread to regional lymph nodes. Moreover, distant metastases are very rare.

Table 1. Clinical and pathological features of 37 cases of laryngeal liposarcoma

	Authors	Age	Sex	Location	WHO Histology	Treatment	Rec	Follow-Up	year
1	Dockerty [26]	33	M	left AEF	Pleo	surgery	Y	DOD	1968
2	Kapur [1]	61	M	Larynx, left PS	Myxoid WD	surgery	Y 3 Rec	DOD within 12 m	1968
3	Miller [3]	43	M	right FVF	Myxoid WD	TL	N	NED 72 m	1975
4	Velek [4]	68	M	right supraglottis	WD	TL	N	NED 80 m	1976
5	Krausen [5]	67	M	Epiglottis, right AEF	PD	SL. pRT	N	DOD 72 m	1977
6	Ferlito [6]	52	M	left FVF+TVF	Pleo	HL, pRT/TL	Y	NED 72 m	1978
7	Tobey [7]	61	M	right vocal cord	PD	SL, Chem	Y	DOD 13 m	1979
8	Gaynor [8]	59	M	right AEF, Epiglottis	MX/RDC	SL	N	NED 36 m	1984
9	Shah [9]	28	M	Epiglottis	MX/RDC	SL	Y	32 m	1984
10	Gaynor [8]	59	M	Epiglottis, AEF	MX/RDC	SL	Y	36 m	1984
11	Narula [10]	45	M	Larynx	MX/RDC	TL	N	DOD 30 m	1985
12	Allsbrook [11]	45	M	left arytenoid	WD	EE/LP×2, EE	Y	48 m	1985
13	Gadomski [12]	28	M	Epiglottis	WD	SE	N	NED 48 m	1986
14	Meis [13]	54	M	right AEF	WD	EE/EE, SL, ND, pRT	Y	NED 58 m	1986
15	Gertner [14]	37	M	left arytenoid	WD	TL, pRT	N	DOD 24 m	1988
16	Wenig [15]	49	M	AEF	WD, 3 cm	SE/SE	Y	NED 72 m	1990
17	Wenig [15]	65	F	Epiglottis	WD, 2×2×1.5 cm	SE/SE	Y	NED 480 m	1990
18	Wenig [15]	57	M	Epiglottis	WD	SE/SE×2, TL	Y	NED 33 m	1990
19	Wenig [15]	68	M	Epiglottis	WD, 3×2×1.5 cm	TL	N	NED 168 m	1990
20	Wenig [15]	55	M	right FVF	WD, 6×3 cm, polyp	SE/SE×4, pRT	Y	NED 120 m	1990
21	Esclamado [16]	34	M	left AEF	WD	SL	N	NED 15 m	1994
22	McCormick [17]	62	M	Larynx	DD	Not doc	Y	Rec 276 m	1994
23	Hurtado [18]	83	F	left TVF	WD	EE	N	NED 24 m	1994
24	Wenig [19]	76	F	Epiglottis	MX/RDC	LP/TL	Y	DND 12 m	1995
25	Wenig [19]	44	M	Epiglottis	MX/RDC	EE/EE, LP	Y	NED 72 m	1995
26	Wenig [19]	56	M	right TVF	Pleo	EE/SE	Y	NED 60 m	1995
27	Wenig [19]	72	M	Epiglottis	WD	EE/EE×7, LP	Y	NED 120 m	1995
28	Wenig [19]	56	M	left AEF	WD	EE/EE×3	Y	NED 108 m	1995
29	Wenig [19]	63	M	right AEF	WD	SL	N	NED 36 m	1995
30	Gal [20]	63	M	Larynx	WD	SE	Not doc	Not doc	1998
31	Mandell [21]	37	M	left arytenoid	WD	EE/EE	N	RD 122 m	1999
32	Mestre de Juan [22]	62	M	right AEF	WD	SL	N	NED 14 m	1999
33	Acharki [23]	50	M	FVF	MX/RDC	TL, pRT (50 Gy)	N	NED 18 m	1999
34	Brauchle [24]	51	M	Epiglottis	WD	LP	N	NED 18 m	2001
35	Yaqoob [25]	60	F	right TVF	MX/RDC	Not doc	Not doc	Not doc	2006
36	Powitzky [2]	63	M	left AEF, PS	DD, NOS	LP	N	NED 12 m	2007
37	Present case	53	M	right AEF, FVF	WD, 2×1 cm	TL	Y	NED 13 m	2014

Abbreviations: AEF, aryepiglottic fold; Chem, chemotherapy; DD, dedifferentiated; DND, dead of causes other than disease; DOD, dead of disease; EE, endoscopic excision; F, female; FVF, false vocal fold; HL, hemilaryngectomy; LP, laleral pharyngetomy; m, months; M, male; MX, mxyoid; ND, neck disseciion; NED, alive with no evidence of disease; Not doc, not documented in report; PD, poor differentiated; Pleo, pleomorphic; Prt, postoperative radiotherapy; PS, pyriform sinus; RDC, round cell; Rec, recurrence; RT, radiation therapy; SE, simple excision; SL, supragiottic laryngectomy; TL, tolal laryngectomy; TVF, true vocal fold; WD, well differentiated; WHO, World Health Organization.

The locally aggressive behavior of the tumor is underscored by its tendency to recur after surgery. Summarize all published cases, 19/37 patients had recurrence (51%) to date, regional nodal metastasis has not been reported, while distant metastases have been reported in 3/37 patients (8.1%) [2]. The high grade tumors have spread to the skin and the bone, causing the patient's death [6, 16].

The recommended treatment for LLS is wide surgical excision [2], i.e. excision of the lesion with a sufficient cuff of surrounding tissues generally free of disease. In fact, the high recurrence rate of laryngeal liposarcomas seems to be related more to treatment than to the histological type [2]. There appears to be no justification for the use of radiotherapy or chemotherapy in these patients, for these adjuvant techniques do not improve the results obtained by complete resection alone. Radiotherapy has been used only occasionally as adjuvant to surgery or for recurrent lesions following surgery [4, 15]. LLS has a better prognosis compared to

their counterparts in extremities or retroperitoneum, for the symptoms of airway obstruction occur early resulting in early medical examination.

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Disclosure of conflict of interest

None.

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