Vascular Leiomyoma of the Larynx: A Rare Entity

Three Case Reports and Literature Review

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Key Words

Vascular leiomyoma · Larynx

Abstract

Background: We conducted a review of the literature to investigate the clinical presentations and histopathologic findings of laryngeal vascular leiomyomas. **Method:** We retrospectively analyzed 24 cases of vascular leiomyomas of the larynx. **Results:** Vascular leiomyoma of the larynx is a very uncommon tumor with only 24 cases reported in the literature. Of the 24 cases, the male/female ratio was 19/5. The patients were from 11 to 78 years old, and nearly half of the patients (11/24) were between the ages of 40 and 60. The tumor occurrence sites varied, with the supraglottic region being most common (14/24), followed by the glottic region (7/24) and the subglottic region (3/24). A common symptom of laryngeal leiomyoma is hoarseness of voice. The tumor is submucosal with dilated vessels on the surface. Histopathologically, the tumor cells are composed of numerous vari-form blood vessels lacking mitosis and well-differentiated smooth muscle bundles. Complete surgical excision is the most commonly recommended treatment. **Conclusions:** Vascular leiomyoma is a benign tumor. External incision effectively prevents hemorrhage during the operation and re-

currence after the operation. A smaller, well-circumscribed laryngeal vascular leiomyoma may be easily amenable to transoral laser resection. Preoperative embolization can be potentially used to benefit the laryngeal transoral resection.

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Introduction

Vascular leiomyoma is an uncommon type of smooth muscle tumor. It is rarely seen in the head and neck region, including the involvement of the larynx. Our department has treated 3 patients with laryngeal vascular leiomyoma since it was established 27 years ago. Clinical images and histopathologic findings of our cases are summarized here.

Case Reports

Case 1

A 53-year-old male patient complaining about hoarseness was admitted to the hospital in September 1997. Both true vocal cords were mobile. Laryngoscopy revealed a smooth red polypoid mass of 2 × 3 mm in size on the left vocal cord (fig. 1). CT of the larynx

Fig. 1. Stroboscopy showing the 3 × 2 mm polypoid mass in the anterior third of the left vocal cord and lack of dilated vessels on the tumor surface.

Fig. 3. Epiglottis vascular leiomyoma: composed of blood vessels and smooth muscle fibers, which are seen surrounding the vessels, and some mucous degeneration was observed, accompanied by infiltrating of lymphocytes and eosinophil cells. HE. ×100.

Fig. 2. Intraoperatively, the mass was about 2 × 3 cm, locating on the right aryepiglottic fold, and was completely removed.

also revealed a same-sized soft tissue mass under the surface of the anterior third of the left true vocal cord.

The tumor was resected using semiconductor laser by suspension microlaryngoscopy. Intraoperative bleeding was minimal. The postoperative period was uneventful. Subsequent histopathology demonstrated a vascular leiomyoma. The patient is disease-free after a follow-up of 9 years.

Case 2

A 47-year-old male patient, who was admitted in August 1998, complained about a 6-year history of foreign body sensation in his throat and new dysphagia for the 2 months prior to admission. Stroboscopy revealed a pink smooth submucosal mass with many dilated vessels in the right aryepiglottic fold. The right true vocal fold could not be clearly visualized. The remainder of his ENT examination was normal.

The patient underwent tracheotomy. Complete excision of the tumor was performed by lateral neck incision (fig. 2). The postoperative course was uneventful. Histopathology demonstrated a vascular leiomyoma.

No recurrence or malignant transformation has been detected during an 8-year follow-up.

Case 3

A 62-year-old male patient, complaining about a globus feeling in the throat, visited a county hospital in April 2003. Physical examination revealed a red mass on the lingual surface of the epiglottis. An indirect laryngoscopy and biopsy of the lesion was performed on the patient, and a complicated significant hemorrhage from the nutrient artery occurred with a total of 300 ml blood loss. After the bleeding had been controlled by an emergency tracheotomy and local hemostasis, the patient was transferred to our department.

Histopathologic report of the biopsy from the county hospital demonstrated a vascular leiomyoma (fig. 3). Another indirect laryngoscopy in our department revealed a 15 × 10 mm mass with no evidence of active bleeding. Three days after hospital transfer, the patient experienced a sudden hemorrhage of about 500 ml, which induced a hemodynamically unstable situation with blood pressures in the range of 97/67 mm Hg.

An emergency surgery by lateral neck incision was performed to remove the tumor and the patient was discharged 2 weeks later without further complications. No recurrence or malignant transformation has been detected during a 3-year follow-up.

Discussion

Vascular leiomyoma arises from the smooth muscle cells of blood vessel walls [1]. The most common sites of origin include the female genital tract, the gastrointestinal system and the pilar arrector muscles of the skin, and the majority of these tumors occur in the extremities. Angiomyoma does not occur as frequently in the head and neck region [2]. Vascular leiomyoma of the larynx has been rarely reported. To our knowledge, there were only 15 cases reported in the Chinese literature and 9 cases in the English literature [1, 3–6]. Among these 24 patients, 19 were male and 5 were female. Their ages ranged from 11 to 78 years, with a mean of 53.2, and nearly half of the patients were between 40 and 60 years old. The pathogenesis of vascular leiomyoma is unclear. An etiological role of estrogen has been suggested in the uterine angiomyoma. Fibroids of the uterus can regress or atrophy following the menopause, indicating a hormonal role in the etiology of such benign smooth muscle tumors. However, estrogens appeared to have little influence on leiomyomas outside the uterus [3]. Instead of a female dominance, we observed a male dominance (19/24) in our present reviewed cases. All 3 patients in our case reports were male.

Among these reports, the most common site of occurrence for laryngeal leiomyoma is the supraglottic region (14/24), followed by the glottic region (7/24) and the subglottic region (3/24). The common symptoms are hoarseness, dyspnea, dysphagia and sensation of a foreign body in the throat.

Five out of 24 patients with laryngeal angiomyoma required urgent tracheotomy at the time of diagnosis, due to the large size of the mass or considerable hemorrhage after biopsy. Indirect laryngoscopy revealed that the tumor is submucosal with dilated vessels on the surface, slow-growing, wine red in color, pedunculated, round, elastic and single with a clear boundary. CT imaging of the neck demonstrated a homogenous soft tissue mass. Enhanced imaging of the lesions was not uniform [6]. The clinical reports from our department suggested to us that once a smooth, wine red mass in the larynx was found by indirect laryngoscopy, no biopsy should be per-

formed. A mass with those characteristics should be considered as a vascular tumor and not a laryngeal carcinoma. Preoperatively, a CT scan or magnetic resonance imaging should be obtained. Enhanced imaging using digital subtraction angiography should also be considered. If biopsy is pursued, it should be done by suspension laryngoscope after careful preoperative evaluation and preparation.

Histopathologically, the tumor cells are composed of numerous variform blood vessels, lacking mitosis and well-differentiated smooth muscle bundles. Whorls of smooth muscle fibers are observed surrounding the blood vessel endothelium, sometimes with mucoid change. Tumors may become malignant when the diameter of the lesion is more than 20 mm with the presence of tumor necrosis. In addition, if 1–4 mitoses per 10 high power fields were observed, we can consider it as a malignant tumor [1]. Malignant transformation of vascular leiomyoma is highly unlikely. Only 2 cases of angiomyoma have been reported to have undergone malignant transformation and both cases involved angiomyoma of the hand [7–8]. No malignant transformation of angiomyoma of the larynx has been reported from the literatures reviewed.

The treatment of laryngeal vascular leiomyoma has not been standardized due to their rarity. Therapeutic decisions have thus far frequently been made empirically from case to case. Among these 24 case reports, the most recommended treatment for laryngeal vascular leiomyomas is the complete surgical excision via endoscopic (7 cases) or external approaches (16 cases) [3–6, 9]. Even though apparently none of the 16 cases that applied external incision reported perioperative complications or subsequent local recurrence, the endoscopic surgery is still favored, since it causes less tissue damage. In the 1980s, the endoscopic excision had a high risk of significant bleeding due to the tremendous vascularity of these tumors as well as the primitive knowledge on transoral resection at the time [5]. In the past 2 decades, however, transoral instrumentation, laser techniques and particularly surgical experience have advanced enormously [10]. Therefore, a smaller, well-circumscribed laryngeal vascular leiomyoma could be easily amenable to transoral laser resection these days as presented in case 1.

Preoperative interventional radiologic embolization technique has proven to be effective at preventing serious intraoperative hemorrhage and other complications for vascular leiomyomas at sites other than the larynx [11–13]. This technique has not been applied in any of the 24 reported cases, though applications in tumors in other

locations of the larynx, such as the angiofibroma [14] or the laryngeal paraganglioma [15–16], have been reported. Preoperative embolization could possibly have benefited these laryngeal cases contemplated for transoral resection. When applying this technique, even if serious intraoperative bleeding was encountered during a transoral operation, surgery can be readily switched to the open approach. Transoral approach for anatomically suitable lesions and avoidance of tracheostomy provide patients with the benefits of rapid recovery of the swallowing

function by leaving the pharyngeal innervation and musculature maximally intact. In addition, favorable results have been reported using transoral laser resection for other vascular lesions of the larynx [17].

Vascular leiomyoma is a benign tumor. Although recurrence of this tumor is rare once treated, 1 case of recurrent angioleiomyoma of the larynx has been reported [1]. Patients should be monitored for recurrence on a periodic basis after surgery.

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