# Recurrent laryngeal leiomyoma

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## ABSTRACT

Leiomyoma–a benign smooth muscle tumor which is most commonly reported in uterus and gastrointestinal tract is rare in the upper aero-digestive tract due to the paucity of smooth muscle in the area. To the best of our knowledge, only 40 cases of leiomyoma occurring in larynx have been reported since 1966 in English literature. We report a case in a 18-year-old lady who presented with recurrent laryngeal leiomyoma which was confirmed by immunohistochemistry. The post-operative course was uneventful.

**Key words**: Desmin, laryngeal leiomyoma, supraglottic tumor

## INTRODUCTION

Leiomyoma, a benign smooth muscle tumor which is most commonly reported in uterus and gastrointestinal tract. They are rare in the upper aero-digestive tract, due to the paucity of smooth muscle in this area. A review of the literature confirms that this kind of neoplasm is extremely rare in the larynx and not many cases are described in the literature so far. The present case highlights the diagnostic histopathological features of the tumor.

## CASE REPORT

A 18-year-old female presented with 3 week old history of hoarseness of voice, dysphagia, and sore throat to outpatient clinic. A patient had similar history 5 months back and was diagnosed clinically to have laryngeal papilloma which was resected and was confirmed as leiomyoma by histopathological examination.

On laryngeal telescopy, there was a solitary fleshy, pink mass in supraglottic area above false cords. Direct laryngoscopy performed revealed a solitary lobulated sessile mass measuring 0.5-1cm in maximum diameter at anterior 1/3rd of left vocal cords and anterior commissure. Under general anesthesia micro laryngoscope was introduced and the tumor was excised completely by micro laryngeal instruments. Tumor was extending even into left ventricle from the left vocal cord and anterior commissure. Post-operative follow up was uneventful.

Specimen was sent to histopathological examination. On gross examination, the tumor was polypoidal in nature and was gray white in appearance measuring 1 cm across, entire tissue was processed. Microscopy showed fragments of tissue lined by pseudostratified columnar epithelium and focal area by stratified squamous epithelium. Subepithelium showed proliferation of smooth muscle type spindle cells, arranged in fascicular and storiform pattern. No evidence of mitotic figures seen in section studied [Figure 1]. Final diagnosis of leiomyoma of larynx simple type was made. Further on immunohistochemistry (IHC) the tumor cells showed positivity for desmin and diagnosis of leiomyoma was confirmed [Figure 2].

Post-operative course was uneventful. Patient had no complaints on follow-up after a month and laryngoscopy revealed no signs of recurrence.

## DISCUSSION

Leiomyoma of larynx is a non-epithelial benign mesenchymal neoplasm which is extremely rare. Most commonly leiomyoma is seen in the organs with abundance of smooth muscle that

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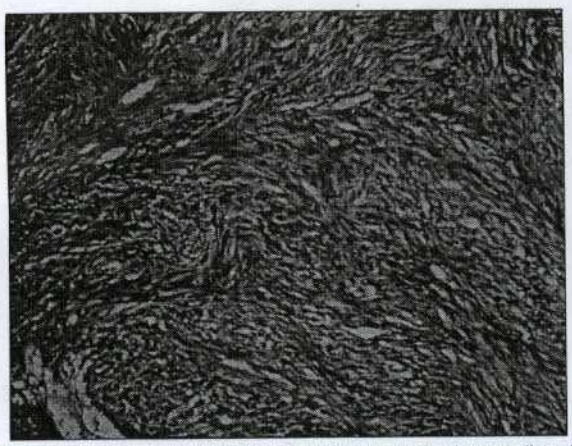

**Figure 1:** Microphotograph of laryngeal leiomyoma showing spindle cells arranged in fascicles and storiform pattern. (H and E, ×100)

**Figure 2:** Microphotograph showing desmin positivity

is uterus and gastrointestinal tract. Cases of leiomyoma seen in other than the uterus and gastrointestinal tract are rare. Leiomyomas located in the larynx are also unusual because there is a little smooth muscle in the larynx compared with the uterus and gastrointestinal tract.[1,2] To the best of our knowledge, only 40 cases of leiomyoma occurring in larynx has been reported since 1966 in English literature.[3]

Leiomyoma of the larynx may be located in the supraglottic, glottis, or subglottic. Most of the reported cases are located in the supraglottic region or glottis.[1] The symptoms of laryngeal leiomyoma include hoarseness, dyspnea, and sensation of a foreign body or pain in the throat. In our case also the patient presented with similar complaints and the tumor was located in supraglottic.

Leiomyoma has been reported in children but more often it has been seen to occur in adults of any age group. There is overall male predominance with male: female ratio of 2:1. The mean age of presentation is 43 years.[3] The present case was a female patient, presented with recurrence at a younger age.

The etiology is unclear. Evidence has shown that the origin of this tumor could be probably from the smooth muscle of the wall of blood vessels, erector pili muscles of hair follicles, or aberrant undifferentiated mesenchyme.[3] Three types of leiomyoma have been described: simple, vascular (angiomyoma), and epithelioid.[1] All three types have been found in the larynx. Vascular leiomyoma, also called as angiomyoma is a rare tumor; The pathogenesis of vascular leiomyoma is unclear and it probably originates from smooth muscle cells within blood vessel walls. It is a slow growing benign tumor that seldom recurs following complete excision. Rich in vascular components it is usually found in the skin of the limbs and head.[1] Histologically, it is an encapsulated tumor and consist of smooth muscle bundles amongst numerous vascular spaces. 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. The differential diagnoses probably thought are hemangioma and vascular leiomyosarcoma.

- Morimoto (1973) described three histologic subtypes:
  - Solid (capillary): smooth muscle bundles surround vascular slit-like channels.
  - Cavernous: dilated vascular channels, less smooth muscle.
  - Venous: vascular channels with thick muscular walls; most common type in the head and neck.
- Malignant variants are rare:
  - Absence of mitosis is the most useful indicator of a benign lesion.[4]

Epithelioid leiomyoma occur mostly in stomach. Here cells are arranged in solid nests and sheets mainly epithelioid, round, or spindle shaped cells with clear cytoplasm. The tumor cells shows positive immunoreactivity for smooth muscle actin and desmin. The tumor shows low mitotic activity and immunostaining with MIB 1 (Ki-67) reveals the occasional cell to be positive, confirming a low proliferative activity in agreement with a benign neoplasm. Epithelioid leiomyomas located in other sites have been shown to act occasionally in a malignant fashion, and the necessity for careful long-term follow-up of the patient is therefore emphasized.[5]

Definite diagnosis of leiomyoma depends on histological examination. The characteristic findings of simple leiomyoma are as follows: interlacing fascicles of spindle-shaped cells, cigar-shaped nuclei, eosinophilic cytoplasm, and a positive reaction to smooth-muscle actin or desmin by IHC.[1,3]

Elevated mitotic activity, anaplasia, and bizarre cell forms are criteria used to differentiate malignant leiomyosarcomas from benign leiomyomas.[1] Our case showed histological features of leiomyoma and IHC positivity for desmin confirmed the diagnosis.

Ultrastructural morphology of the tumor shows basement membrane enveloped leiomyoma cells containing variable numbers of pinocytotic vesicles in close proximity to the plasma membrane. Heavy concentration of microfilaments, dens bodies, and marginal dens plaque are seen in the cytoplasm.[6]

These lesions have a very low rate of recurrence or malignant degeneration. But in this case patient had recurrence of the tumor within a period of 5 months.

## CONCLUSION

Laryngeal leiomyoma is a very infrequent neoplasm and should be kept in the mind of histopathologist while evaluating the tumors of upper aero-digestive tract. In our case the unusual features are tumor seen in young female patient with recurrence.

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