CASE REPORT

Worm in the Throat: Hamartoma Larynx

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ABSTRACT

Hamartomas of larynx are rare benign lesions which can be a rare cause for respiratory obstruction. This report highlights the case of a young female who presented with foreign body sensation and worm-like mass in throat of 10 years duration. On videolaryngoscopy, a pinkish polypoidal worm like mass was seen arising from the right side of epiglottis on the laryngeal surface. Endoscopic guided excision with cautery was done and the mass revealed hamartoma on histopathology.

Keywords: Hamartoma, Videolaryngoscopy, Epiglottis.

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INTRODUCTION

Albrecht introduced the term 'hamartoma' in 1904,¹ and distinguished between true neoplasms and tumor like lesions. In 1991, the World Health Organization defined hamartomas as anomalies characterized by the formation of a tumor like mass composed of identical mature cellular tissue elements that are normally present where the mass is found but that occur in abnormal proportions or patterns.² It may arise from any of the germ layers and does not metastasize. Although hamartomas have been described as occurring throughout the body, they remain a rare finding in the head and neck region. Hamartoma may present as a single lesion or in terms of a multiple hamartoma syndrome.^{3,4} They usually involve the lungs or organs in the abdominal cavity. The lesion usually presents as a submucosal mass with ill-defined margins. Microscopically, the tissue shows mesenchymal elements, either in isolation or mixed loosely with epithelial structures in a disorganized architectural pattern. Hamartomas are

identified according to the preponderance of a particular structure (cartilage, fat, neural structures or fibromuscular tissue). Mesenchymal hamartomas, which contain only mesodermal elements, are much more common than epithelial or glandular hamartomas, which show a mixture with mesodermal element.⁵

We report a case with the unusual presentation of worm-like mass in throat that was consistent with laryngeal hamartoma. The clinicopathologic features and treatment, are discussed along with review of relevant literature.

CASE REPORT

A 27-year-old female presented to our outpatient department with history of foreign body sensation and worm like mass in the throat (Fig. 1) since 10 years. There was difficulty in breathing since 3 days prior to admission. On videolaryngoscopy, a smooth, shiny elongated pinkish worm-like mass was seen arising from the right side of the epiglottis on its laryngeal surface. The mass was freely mobile at one end and attached to the epiglottis at the other end (Fig. 2). The examination of nose and ears was normal. A provisional diagnosis of hamartomatous polyp and leiomyoma was thought of.

The case was posted for endoscopic guided excision of the mass under general anesthesia with number 6 endotracheal tube. The worm like mass (Fig. 3) was excised with cautery from right side of the laryngeal surface of the epiglottis. The mass was sent for histopathological examination (Fig. 4) which revealed, hyperplastic stratified squamous epithelium, smooth muscle bundle zone, seromucinous gland zone, along with duct and vascular zone, suggestive of hamartomatous polyp. The immediate postoperative period was uneventful. At the first postoperative visit, 1 week after surgery, the patient's symptoms had resolved. The surgical site had healed. There has been no evidence of recurrent lesion till the last follow-up of 3 years.

DISCUSSION

Hamartomas are benign lesions, most commonly found in the lungs, kidneys and intestine. Although they are rarely seen in head and neck, they can develop in nasopharynx, nasal cavity, eustachian tube, tongue, oropharynx, hypopharynx, larynx, cervical esophagus and trachea.^{6,7}

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Fig. 1: Worm-like mass in oral cavity

Fig. 2: Preoperative videolaryngoscopy

Fig. 3: Tumor resected *en bloc* hamartomatous polyp

Fig. 4: HPE showed a central core of dilated vascular spaces surrounded by a matrix of fibrous connective and adipose tissue covered by normal squamous epithelium, with no evidence of hyperplastic or dysplastic changes

The pathophysiologic mechanism underlying the origin of laryngeal hamartomatous polyps is unknown. In this case, the origin of the hamartomatous polyp from laryngeal surface of epiglottis cannot be explained by muscular weakness.⁸ Nonetheless, a small mucosal lesion in that region exposed to the pressure changes associated with deglutition and innumerable boluses of swallowed material could provide a nidus for the formation of a giant hamartomatous polyp.

The first report of a laryngeal hamartoma was published by Climie et al⁹ in 1963. Zapf et al¹⁰ in 1981, described a supraglottic polypoid lesion in a 6-week-old child that caused increasing stridor and sternal retraction. In infants, laryngeal hamartomas are typically associated with severe laryngeal obstruction.¹⁰⁻¹² In adults, the signs and symptoms of laryngeal hamartomas are more diverse. Some adults experience hoarseness, dyspnea, and even an acute airway obstruction that may require tracheotomy.^{7,13} Other adults experience a longstand-

ing and sometimes nearly asymptomatic course of disease.⁹ Our patient did not notice any laryngeal symptoms for many years of onset, despite her slight but noticeable hoarseness to the examiner. Patient noticed recurrent episodes of dyspnea and choking sensation prior to admission after prolonged asymptomatic period.

Any laryngeal tumor has to be investigated by video laryngoscopy and computed tomography scan to assess the extent of the tumor.¹⁴ Although it has been shown that most laryngeal hamartomas develop in the supraglottic region as in our case, retrolaryngeal tumor growth and coincidence with laryngeal clefts¹⁵ has to be excluded by meticulous endoscopy of the larynx and hypopharynx. Final diagnosis is based on histopathological examination. If the diagnosis is established, total resection of the lesion is the treatment of choice. In present case, on video laryngoscopy, a smooth, shiny elongated pinkish worm like mass with base attached to the

right laryngeal surface of the epiglottis with freely moving at other end was noted.

The mass was subjected for total excision under general anesthesia with endoscopic assisted cautery and sent for frozen section which revealed hamartomatous polyp. Post-operative period was uneventful and there was no recurrence of tumor after 3 years of follow-up. If the lesion is well-encapsulated and of small size on presentation, endoscopic removal using a CO₂ laser/cautery, either alone¹⁶ or in combination with microscopic instrument,¹⁷ is adequate. Zapf et al¹⁰ debulked a supraglottic polypoid lesion with a snare through a rigid endoscope. Postoperative complications after endoscopic surgery (i.e. immediate massive hemorrhage) have been reported.¹⁸ An open approach should be indicated for larger tumor.^{5,10} A transpharyngeal approach will expose retrolaryngeal extent of tumor growth.

CONCLUSION

Our case is rare variety of presentation in view of long history of nontroubling foreign body sensation in the throat with recent history of breathing difficulty and worm like mass in the throat. When symptoms are vague, laryngeal hamartomas may be misdiagnosed as a psychiatric disorder.

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