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Case report/Kazuistyka

Intravascular papillary endothelial hyperplasia of larynx: Case report and literature review of all head and neck cases

Miroslav Tedla [1,2,*], *Miroslava Bežová* [1,3], *Csaba Biró* [4], *Eva Tedlová* [5], *Chee-Yean Eng* [6], *Karol Zeleník* [7]

[1] *University ENT Department, Medical School of Comenius University and University Hospital Bratislava, Slovakia*
[2] *ENT Department, University Hospitals Coventry and Warwickshire, University of Warwick, United Kingdom*
[3] *ENT Department, Faculty Hospital Trenčín, Slovakia*
[4] *Histopatológia a.s., Bratislava, Slovakia*
[5] *Department of Pneumology and Phtiseology, Medical School of Comenius University and University Hospital Bratislava, Slovakia*
[6] *ENT Department, Alexandra Hospital, Singapore*
[7] *Department of Otolaryngology, University Hospital Ostrava, Czech Republic*

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ABSTRACT

Intravascular papillary endothelial hyperplasia (IPEH) is a benign lesion of a vascular origin that is caused by excessive proliferation of endothelial cells in blood vessels or vascular malformations. It is a rare entity that can present in any region of the body, but with particular predilection to the head and neck region and the extremities. We also present the results of the English literature search, which to our knowledge are all the published cases of IPEH in the head and neck region (No = 213). IPEH has not been reported to arise from the glottic region previously. We present a first case of IPEH arising from the vocal fold of a 48-year-old male. Histological differential diagnosis of IPEH includes several entities, most importantly angiosarcoma. Presentation and histology are discussed. The main treatment option is a complete surgical resection. Prognosis of IPEH is excellent, with the exception of some intracranial cases.

Introduction

Abnormal tissue in the larynx poses a clinical dilemma as chronic dysplastic/hyperplastic changes or squamous-cell carcinoma (SCC) is not rare in middle age male smokers. Numerous benign pathologies may present, for instance: nodules, polyps, contact granulomas, and papillomas. One difficult-to-diagnose entity is intravascular papillary endothelial hyperplasia (IPEH), also known as intravascular

* *Corresponding author at*: University Hospitals Coventry and Warwickshire, Clifford Bridge Road, Coventry CV22DX, United Kingdom. Tel.: +44 247 696 56 84; fax: +44 247 696 56 13.
E-mail address: miro.tedla@gmail.com (M. Tedla).
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Table I – Published cases of IPEH in the larynx		
Present report	1	Vocal fold
Guvenc 2008	1	Hypopharynx and larynx
Sezgin 2005	1	Supraglottic larynx

angiomatosis, Masson's pseudoangiosarcoma, Masson's tumor or vegetant intravascular hemangioendothelioma [1]. IPEH is a benign lesion of vascular origin that is caused by an extensive proliferation of endothelial cells in normal blood vessels or vascular malformations. Although ubiquitous in nature, IPEH has a predilection for the head and neck [2].

An English language literature review was performed. Pubmed search engine was used with the phrase ''intravascular papillary endothelial hyperplasia'' or ''Masson's tumor'' in the title or/and abstract. Reference search has been performed to identify further articles of interest. One hundred and 76 articles were identified. After manual narrowing of our search to the Head and Neck region, 68 reports of Head and Neck IPEH were identified. A total of 213 cases of IPEH pertaining to the Head and Neck region have been identified. We present a review of sites of IPEH published in the literature so far in the larynx (Table I).

Most of the reported cases in the head and neck have involved the oral mucosa, skin, subcutaneous tissue of the face and scalp, intracranium, orbit and ocular adnexa. We identified two reports of IPEH in the larynx: an 18-year-old female with a mass in the hypopharynx and larynx [3] and extravascular IPEH of the larynx in the mucosa of the epiglottis extending to the anterior commissure [4].

In this article we publish the first case of laryngeal IPEH at the level of the vocal folds. To our best knowledge it is only the third published case of IPEH affecting the larynx.

Case description

A 48-year-old male presented to the regional ENT unit with a three-year history of hoarseness. He had no history of cough, dysphagia, bleeding or upper respiratory tract infections. He had a history of smoking 25 packs/year and was a moderate alcohol drinker. His medical history was unremarkable. Laryngoscopy revealed a large oval tumor with a smooth surface growing from the inferior aspect of the left vocal fold. CT showed a polypoid tumor sized 17 mm × 11 mm × 8 mm (AP, LL, CC) with prominence into the laryngeal cavity (Fig. 1). The origin of the tumor was at the level of thyroid cartilage (level of C6). The density of tissues was 30–45 HU, which showed vascular areas and vessels. No invasive growth or lymphadenopathy was identified. Any possible invasion from the adjacent structures in the neck was excluded by an MRI. Microlaryngoscopy under general anesthesia was performed and a flaccid tumor, sized 12 mm, was completely excised from the inferior aspect of the left vocal fold. Histology demonstrated IPEH (Fig. 2). Immunohistochemical studies revealed a diffuse staining reaction for the presence of Factor VIII-related antigen. A follow-up appointment after one month showed 3 mm residual tissue at the edge of the left fold, and the patient was referred to our tertiary care ENT department. During a repeat microlaryngoscopy this residual tumor was excised. Macroscopically it

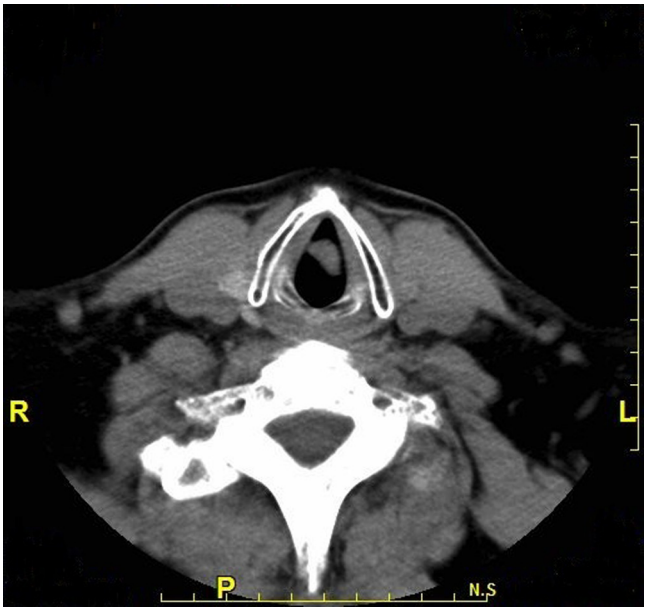

Fig. 1 – IPEH – CT-axial scan at the level of vocal folds

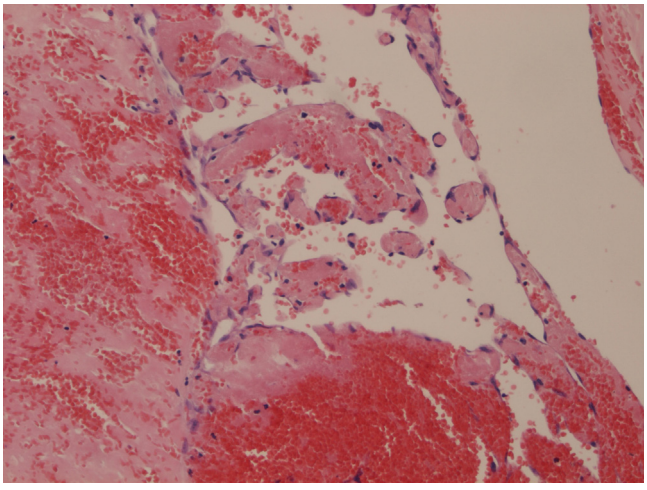

Fig. 2 – Histopathology – vascular wall with IPEH (hematoxylin & eosin, 200×)

appeared as hemangioma, histologically confirmed as IPEH. Biopsies of the margins were free of tumor. At two follow-up visits (after 2 weeks and 8 months) smooth tumor free folds have been observed and the patient was free of symptoms.

Discussion

Presentation

IPEH is a relatively rare lesion occurring in the veins of the dermis and subcutis of the fingers, head and neck, trunk, lower extremities and upper extremities in the order of decreasing frequency. IPEH usually presents as a slow-growing nodule/mass that may be somewhat painful. If

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