CASE REPORT

Recurrent Epistaxis: The First Hemorrhagic Chondro-osseous Respiratory Epithelial Adenomatoid Hamartoma (COREAH) Reported in the Literature

David Temmermand¹, Andriy Pavlenko², Tasos Aslanidis³

ABSTRACT

Chondro-osseous respiratory epithelial adenomatoid hamartomas (COREAHs) are benign entities that are rarely present within the nasal cavity but to date have only been identified in nasal polyp-like masses. This report represents the first case published with a COREAH causing epistaxis. Although these are benign lesions which are nearly 100% curable with simple surgical excisions, they remain rare and are not commonly thought of in the differential of nasal masses or as hemorrhagic lesions. Further, the patient reported here is the oldest reported and has a unique presentation.

Keywords: Endoscopic sinus surgery, Epistaxis, Nasal mass, Nasal polyp.

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INTRODUCTION

Chondro-osseous respiratory epithelial adenomatoid hamartomas (COREAHs) are benign entities that are rarely present within the nasal cavity. Having only been described in the pathology literature around 30 years ago, there are just 7 cases described in the literature to date. And unlike the current case, all have presented as polyp-like masses causing nasal obstruction. Here we report the first case to present as a hemorrhagic lesion along the anterior septum. Classic examples of hemorrhagic lesions of the nasal septum can be hemangiomas (most commonly capillary type on septum),¹ pyogenic granulomas, carcinoma, telangiectasias (hereditary or spontaneous), or hemorrhagic polyps. Certainly, this list is not exhaustive but has not historically included COREAHs.

Our preoperative diagnosis was suspicious for a hemangioma vs pyogenic granuloma. The patient was treated conservatively in the office with multiple rounds of cautery until ultimately requiring complete surgical excision. Fortunately, surgical excision is agreed upon as the appropriate treatment modality for all of these lesions. Recurrence in the nasal cavity remains low (only 1 case demonstrated recurrence and may be attributed to incomplete margins during initial surgery due to its difficult location).² Because of their benign nature, no further care is needed.

CASE DESCRIPTION

Mr A is 75-year-old male with a several-year history of left epistaxis and no history of allergies. Approximately 1 month prior to surgery, the patient had a 3 mm raised lesion in Little's area of the left septum. He was cauterized in the clinic with silver nitrate multiple times over multiple days (Fig. 1). Over the subsequent 2–3 weeks, the patient demonstrated continued episodes of heavy left sided epistaxis described as pulsatile at times. Conservative management of pressure and oxymetazoline soaked cotton balls failed to control the bleeding and ultimately required a balloon-based packing (RapidRhino). On the day of surgery, there was a non-friable, irregular, white scar-like area on the left anterior/inferior nasal septum without significant ectasia of surrounding vessels. The 3 mm

¹Department of Otolaryngology-Head and Neck Surgery, Augusta University, Augusta, Georgia, USA

²Department of Pathology, Kennedy University Hospital, Stratford, New Jersey, USA

³Department of Otolaryngology-Head and Neck Surgery, Rowan University School of Osteopathic Medicine, Blackwood, USA

Corresponding Author: David Temmermand, Department of Otolaryngology-Head and Neck Surgery, Augusta University, Augusta, Georgia, USA, Phone: +1 7067216100, e-mail: temmerda@rowan.edu

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Fig. 1: Submucosal glandular proliferation with respiratory epithelium-lined mucosa, hypocellular fibromyxoid stroma, and mature bone (hematoxylin and eosin, 40x)

Fig. 2: Hematoxylin and eosin, 100×

area was excised down to perichondrium with the defect allowed to heal secondarily. At the 3-month follow-up, the septum has healed without incident and he has had no additional epistaxis (Fig. 2).

DISCUSSION

Hamartomas are benign neoplasms composed of abnormal mixtures of mature cells indigenous to the local tissues with an excess of one or more cell types and can be further classified as epithelial, mesenchymal, or mixed epithelial and mesenchymal types.^{3,4} Within the nasal cavity, respiratory epithelial adenomatoid hamartomas (REAH) are far more common than the chondro-osseous respiratory epithelial adenomatoid hamartoma (COREAH).

The etiology of COREAHs are unknown although they are histologically similar to inflammatory polyps: stromal edema, polypoid growth, increased numbers of sero-mucinous glands, vascular and fibroblastic proliferation, and mixed acute and chronic inflammatory cells.^{2,3,5} Interestingly, our case was not polypoid, but given prior cautery he may have been caught at an early stage before a large polyp could have been identified.

Some also believe that REAH/COREAHs are similar to other hamartomas, which is to say that they are congenital in nature.^{3,4} This is unlikely to be true given the broad age spectrum this disease presents. The previously reported age range is from 3 to 68 (3, 7, 11, 38, 34, 68, 59, and now 75).

Most of the cases have been documented from the turbinates, sinuses, lateral walls, or nasal roofs. Only one was previously documented from the nasal septum but originated posteriorly. Further, all were found during cases of polyp resection secondary to nasal obstruction.^{6,7} This is the first case described as a nonpolypoid septal lesion and the first to present as recurrent epistaxis.

Treatment remains straightforward as simple surgical excision is enough to provide near total cure rates.⁵ The learning focus for this case is an expansion of the differential for hemorrhagic lesions within the nasal cavity and the ability of surgical excision to resolve this patient's epistaxis.

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