Case Report

DOI: http://dx.doi.org/10.18203/issn.2454-5929.ijohns20201701

Respiratory epithelial adenomatoid hamartoma presenting as a mass in nasal cavity: a case report

Visweswara Rao Suraneni, Vineela Gaddipati*, Anusha Allena

Department of ENT, Maharajah's Institute of Medical Sciences, Nellimarla, Vizianagaram, Andhra Pradesh, India

Received: 21 January 2020 Revised: 10 March 2020 Accepted: 12 March 2020

*Correspondence:

Dr. Vineela Gaddipati,

E-mail: vineela.gaddipati@gmail.com

Copyright: © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

This article aims to report a rare case of respiratory epithelial adenomatoid hamartoma (REAH) arising from nasal septum that initially presented as bilateral sino nasal polyposis. A 45 years old male presented with a complaint of bilateral nasal obstruction for the past 3 years which was progressive and was planned for functional endoscopic sinus surgery. Polypoidal tissue was found to be coming from nasal septum intraoperatively. Histopathological examination confirmed the presence of REAH. This case demonstrates the rare presentation of REAH, clinically presented as sino nasal polyposis and confirmed as REAH by histopathological examination.

Keywords: REAH, Sino nasal polyposis, Nasal septum, Histopathological examination

INTRODUCTION

Hamartoma is defined as an aberrant differentiation which may produce a mass of disorganized, mature specialized cells or tissue indigenous to the particular site.¹

Hamartomas are first described in 1904, can occur in any area of the body, but commonly originate from the spleen, lung and intestine.² In 1995, Birt and Knight-Jones reported the first case of a hamartoma in the upper aerodigestive tract.³

In 1995, Wenig and Heffner and introduced the term respiratory epithelial adenomatoid hamartoma (REAH) as one particular type of hamartoma. Approximately 70% of REAHs occur in the nasal cavity, most often localised to the posterior part of the nasal septum.⁴ The lesions also have been reported to occur in paranasal sinuses and nasopharynx.^{5,6}

CASE REPORT

A 45 years old male presented with bilateral nasal obstruction more on left side for 3 years which was insidious in onset and gradually progressive in nature, aggravated with upper respiratory tract infections and relieved with medication. There is history of clear mucoid nasal discharge, on and off episodes which aggravated on exposure to dust and cold weather, relieved with medication.

Anterior rhinoscopy on both sides showed polypoid masses which are pale, with regular surface, soft in consistency, non tender, not sensitive to touch, no bleed on touch. Diagnostic nasal endoscopy was done in Figure 1a and 1b.

Computed tomography (CT) scan of paranasal sinuses showed opacities in the nasal cavity and bilateral maxillary sinuses in Figure 2a and 2b.

Intraoperatively, under endoscopic guidance the polypoid mass on left side was debrided and found to be coming from nasal septum. The mass was excised and sent for histopathological examination.

Histopathological examination revealed polypoid tissue lined by pseudostratified columnar epithelium. Stroma is edematous enclosing several cystic spaces lined by columnar to mucinous epithelium. Several inflammatory cells comprising predominantly of eosinoplils are seen. No cellular atypical features are seen giving an impression of respiratory epithelial adenomatoid hamartoma (REAH) in Figure 3a and 3b. The patient was followed up to 1 year, no recurrence was observed.



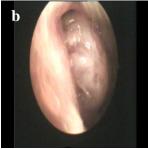


Figure 1 (a and b): Endoscopic images showing polypoidal mass in both right and left nasal cavities.

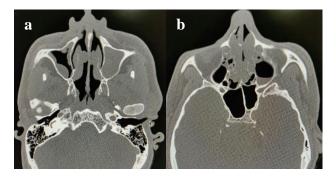


Figure 2 (a and b): CT-PNS showing opacities in bilatera nasal cavities and maxillary sinuses.

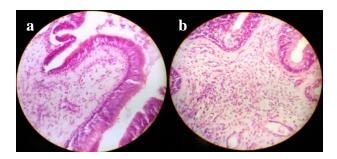


Figure 3 (a and b): Microscopic images showing respiratory epithelium with submucosal glandular proliferation, polypoid changes and inflammatory cells.

DISCUSSION

Respiratory epithelial adenomatoid hamartoma is very rare in the sino nasal tract and was first recognized as a distinctive entity by Wenig and Heffner in 1995.⁴ Symptoms of REAH varied depending on the location of the lesions and included nasal obstruction, rhinorrhea, headache, hyposmia, anosmia, epistaxis, facial pain, proptosis.

REAH has clinical presentations, histological changes, treatment, and behaviour similar to inflammatory polyps, suggesting that the development of REAH is secondary to inflammatory process.² CT and magnetic resonance imaging shows no characteristic signals because they vary according to the main element of the Hamartoma.⁷ Radiologically, the most common finding of REAH is an opacification of the affected sinus and some connection to nasal septum. The lesion also tends to grow slowly, as evidenced by bony expansion rather than erosion.⁸

On gross evaluation, REAHs appear as edematous-appearing polypoid masses. Histologically, the principal morphological aspect is a proliferation and accumulation of glands and ducts covered by pseudostratified, ciliated epithelial cells with no metaplastic or atypical changes. The invagination of the respiratory epithelium leading to the glands may be observed. The characteristic glandular components consist of respiratory epithelium originating from the surface epithelium.⁹ Simple excision is the only treatment as no malignant potential is recognized for REAH.¹⁰ Because of the limited growth potential of a hamartoma, recurrence is very rare.¹¹

CONCLUSION

REAH is an uncommon benign entity with distinctive histologic features. Nowadays it becoming prevalent due to awareness to clinicians with number of reported cases. A misdiagnosis of REAH as inflammatory nasal polyp, low grade adenocarcinoma, inverted papilloma or other granulomatous conditions (e.g. rhinosporidiosis) would likely to result in more aggressive surgical intervention than needed. Therefore, it is important to differentiate it from other pathologies.

ACKNOWLEDGEMENTS

I am thankful to the department of pathology for their support.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

REFERENCES

- 1. Kumar V, Abbas AK, Fausto N. Neoplasia. Robbins and Cotran pathologic basis of human disease, 7th edition. Philadelphia, PA: Elsevier-Saunders Publishing Company; 2005: 272.
- Delbrouck C, Fernandez Aguilar S, Choufani G, Hassid S. Respiratory epithelial adenomatoid

- hamartoma associated with nasal polyposis. Am J Otolaryngol. 2004;25(4):282-4.
- 3. Birt BD, Jones KEB. Respiratory distress due to naso-pharyngeal hamartoma. Br Med J. 1969;3(5665):281-2.
- 4. Wenig BM, Heffner DK. Respiratory epithelial adenomatoid hamartomas of the sino nasal tract and nasopharynx; a clinico-pathologic study of 31 cases. Ann Otol Rhinol Laryngol. 1995;104(8):639-45.
- 5. Himi Y, Yoshizaki T, Sato K, Furukawa M. Respiratory epithelial adenomatoid hamartoma of the maxillary sinus. J Laryngol Otol. 2002;116(4):317-8.
- 6. Metselaar RM, Stel HV, Baan S. Respiratory epithelial adenomatoid hamartoma in the nasopharynx. J Laryngol Otol. 2005;119(6):476-8.
- 7. Kaneko C, Inokuchi A, Kimitsuki T, Kumamoto Y, Shinokuma A, Natori Y, et al. Huge hamartoma with inverted papilloma in the nasal cavity. Eur Arch Otolaryngol. 1999;256(1):337.
- 8. Athre R Ducic Y. Frontal sinus hamartomas. Am J Otolaryngol. 2005;26:419-21.

- 9. Endo R, Matsuda H, Takahashi M, Hara M, Inaba H, Tsukuda M. Respiratory epithelial adenomatoid hamartoma in the nasal cavity. Acta otolaryngol. 2002;122(4):398-400.
- 10. Ozolek JA, Hunt JL. Tumor suppressor gene alterations in respiratory epithelial adenomatoid hamartoma (REAH): comparision to sinonasal adenocarcinoma and inflamed sino nasal mucosa. Am J Surg Pathol. 2006;30(12):1576-80.
- 11. Flavin R, Russell J, Phelan E, Dermott MB. Chondro-osseous respiratory epithelial adenomatoid hamartoma of the nasal cavity: a case report. Int J Pediatr Otorhinolaryngol. 2005;69(1):87-91.

Cite this article as: Suraneni VR, Gaddipati V, Allena A. Respiratory epithelial adenomatoid hamartoma presenting as a mass in nasal cavity: a case report. Int J Otorhinolaryngol Head Neck Surg 2020;6:992-4.