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A rare case of a periorbital respiratory (choristomatous) cyst

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Abstract

Respiratory cysts are benign lesions lined by normal respiratory epithelium. There are few reported cases localized to the orbit, while those of the eyelid are exceedingly rare. Respiratory cysts usually arise either from a non-hereditary congenital malformation, where they are distinguished as choristomatous, or from trauma. Here, we report a case of a 53-year-old man who presented with a large right lower eyelid cyst that was histopathologically diagnosed as a respiratory cyst.

Introduction

Respiratory cysts can be either congenital (choristomatous) or acquired. Congenital cysts are thought to originate from respiratory epithelium sequestered during intrauterine development of the paranasal sinuses.¹ In cases of acquired cysts, they are considered to arise from the displacement of respiratory epithelium due to disruption of normal anatomical barriers, possibly stemming from trauma.² In addition, it is thought that these cysts can also arise from chronic sinus disease complicated by mucocoele formation.³ Here, we report a case of a benign cyst with features of a respiratory cyst of the eyelid with no history of trauma or sinus disease.

Case Report

A 53-year-old man presented to the dermatology clinic with a 1 cm cyst of his lower right eyelid. He reported that the cyst had been present for over a year, during which time it had grown in size. The cyst was soft and mobile, located medially and just inferior to the lash line, distorting the contour of his lower right eyelid. The patient denied any discomfort but complained that it was obstructing his vision.

His medical history was negative for prior orbital and paranasal sinus trauma. However, he endorsed the use of cocaine two times per week. Otherwise, the rest of his clinical history was unremarkable. On ocular exam, his pupils and extraocular movements were unremarkable except for the lesion of the lower right eyelid. In addition, his oropharynx and nasal mucosa were normal.

The cyst was completely excised under local anesthesia. It partially involved the orbicularis oculi muscle but was anterior to tarsus. Histopathological examination revealed a large cystic lesion in the stroma, lined by ciliated columnar epithelium with focal mucin vacuoles (Figure 1). The cyst contained cellular debris and inflammatory cells. There was focal inflammation surrounding the cyst, involving the epithelium, suggesting focal rupture. Multiple step sections were obtained and no other germ cell components were found in the cyst wall.

Discussion

The most common cysts of the periorbital and orbital area are hidrocystomas, epidermoid cysts, and sebaceous cysts; the most common choristomatous cysts include dermoid and epidermoid cysts. Cysts can present with eyelid swelling, proptosis, or strabismus.⁴ However, periorbital respiratory cysts are very rare. The lesion was initially diagnosed as a sebaceous cyst due to its clinical appearance and location. It is important to consider that post-traumatic cysts may present many years after trauma, and become secondarily infected.⁵ There is only one other reported case of a respiratory choristomatous cyst occurring in the eyelid by Jakobiec et al. Similarly, their patient presented without any symptoms or history of sinus disease or trauma. However, the cyst was restricted to the upper eyelid while our patient's cyst occupied the lower eyelid.⁴ Morris et al. documented two cases of respiratory cysts located in the temporal region of the orbit without involvement of the paranasal sinuses.⁶ Similar to our case and Jakobiec et al., the patients did not present with any external inflammatory signs and had no history of orbital trauma or surgery as well.^{4,6} In the present case, our patient presented with at least a yearlong history of a right lower eyelid lesion. He denied a history of chronic sinus disease and orbital trauma and had no history of prior orbital surgery. Additionally, he had no external inflammatory signs despite the presence of inflammation histopathologically.

Microscopically, the lesion was lined by densely packed pseudostratified columnar epithelium with cilia and mucin vacuoles. Our findings are similar to prior descriptions; however, the density of ciliated and goblet cells may vary. For instance, Jakobiec et al. noted high density of goblet cells in the epithelium of their cyst while we observed more intracellular mucinous vacuoles. In addition, these cysts can also exhibit other types of cells such as cuboidal.⁶ Though our lesion possessed clearly respiratory epithelium, acid-fast and phosphotungstic acid staining, as well as immunostaining for CK17 and CK18, can facilitate the identification of cilia or respiratory epithelial lineages, respectively, when cells are not clearly identifiable.⁴ It is important to note that the presence of ciliated cells is not exclusive to respiratory cysts. Therefore, a thorough examination for other germ cell elements is necessary to exclude other entities, such as a teratoma.

It has been hypothesized that acquired lesions may originate from an anterior ethmoidal rudiment that grows slowly over time.⁴ Given that our lesion was localized to the lower eyelid, ectopic tissue from the lacrimal drainage system should be considered. However, the cyst was not seen to involve the punctum clinically, and intraoperative inspection did not reveal any connection or adhesions to the lacrimal sac. Lastly, a breach in the antero-medial orbital wall should also be deliberated given the history of cocaine use. Midfacial

destruction caused by cocaine use has been well-documented in the literature but the coexistence of a respiratory cyst with cocaine use has not been reported to our knowledge.⁷ It is possible that necrosis and perforation of midfacial structures due to cocaine abuse can facilitate the development of an ectopic respiratory cyst by disrupting normal anatomic barriers.

Treatment for cystic lesions vary depending on patient symptoms. Often, surgical removal is recommended to alleviate burdensome symptoms such as vision obstruction. Complete excision is often curative and necessary to avoid future recurrence.^{4,5} Though rare, respiratory (choristomatous) cyst should be considered in the differential diagnosis of eyelid lesions, even in patients who lack a history of trauma or chronic sinus disease.

References

1. Newton C, Dutton JJ, Klintworth GK. A respiratory epithelial choristomatous cyst of the orbit. *Ophthalmology* 1985; 92(12): 1754–1757. [PubMed: 4088630]
2. Mee JJ, McNab AA, McKelvie P. Respiratory epithelial orbital cysts. *Clin Exp Ophthalmol* 2002; 30(5): 356–360. [PubMed: 12213161]
3. Thanos A, Jakobiec FA, Mendoza PR, Hatton MP. Ectopic (choristomatous) orbital respiratory cyst: histopathology and immunohistochemistry. *Surv Ophthalmol* 2014; 59(3): 328–333. [PubMed: 24359804]
4. Jakobiec FA, Roh M, Stagner AM, Yoon MK. Choristomatous respiratory cyst restricted to the upper eyelid. *Ophthalmic Plast Reconstr Surg* 2016; 32(1): e15–e18. [PubMed: 25675165]
5. Goh RL, Hardy TG, Williams RA, McNab AA. Respiratory epithelial cysts of the orbit. *Orbit* 2016; 35(5): 245–249. [PubMed: 27468088]
6. Morris WR, Fleming JC. Respiratory choristomatous cysts in the temporal orbit. *Ophthalmic Plast Reconstr Surg* 2001; 17(6): 462–464. [PubMed: 11766031]
7. Villa PD. Midfacial complications of prolonged cocaine snorting. *J Can Dent Assoc* 1999; 65(4): 218–223. [PubMed: 10224723]

Fig 1:
Photomicrograph of respiratory choristomatous cyst. A. At low magnification, a well-circumscribed cystic lesion is seen, containing eosinophilic debris. B. Cyst is lined by a thin epithelium, which on higher magnification (C) exhibits ciliated columnar morphology with intracellular mucin. D. Close up of ciliated columnar cells, which are consistent with respiratory epithelium. (H&E; A. 15X, stitched image B. 100X C. 200X D. 400X)