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Rare Case of Nasal Vault Oncocytic Cystadenoma

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Rare case of nasal vault oncocytic cystadenoma

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A B S T R A C T

Background: Oncocytic Cystadenomas are a rare benign pathology often found to arise from the salivary glands, reported more commonly in minor salivary glands, but even more rarely in major salivary glands and the larynx. This is the first known report of a nasal vault oncocytic cystadenoma in a pediatric patient.
Methods: Case Report and Literature Review.
Case presentation: A 10-year-old female presented with a mass involving the left nasal vestibule that caused nasal obstruction and a visible external abnormality. MRI imaging revealed a cystic lesion in the anterior nasal cavity that was rim enhancing with a bright T2 signal that appeared to arise from the nasal mucosa. She was taken to the operating room for endoscopic-assisted removal of the lesion. Pathology showed an oncocytic cystadenoma. On follow-up clinic visit, she is doing well with no signs or symptoms of recurrence.
Conclusion: The authors present the first reported case of a pediatric patient with nasal vault oncocytic cystadenoma. In other head and neck locations, these cysts are generally benign and only cause symptoms related to their location and proximity to other vital structures. Surgical endoscopic management was effective for resection.

1. Introduction

Oncocytic papillary cystadenoma is a rare benign tumor of the salivary glands that is generally seen in the minor salivary glands, but can occasionally be seen in the major salivary glands and the larynx [1]. Oncocytes are large, irregularly shaped cells with granular eosinophic cytoplasm that are rich in mitochondria [2]. These cysts are generally asymptomatic unless they progress causing obstruction with symptoms related to their anatomic location and proximity to other structures. In salivary glands, they generally present as painless masses and they have been detected in the larynx causing hoarseness or dysphonia [3]. This report is the first to our knowledge in a pediatric patient with a painless cyst of the nasal vestibule for which histopathology showed an oncocytic cystadenoma. IRB exemption at our institution is permitted for single case reports.

2. Case Presentation

We present a case of a 10-year-old female with history of seasonal allergies and exercise induced asthma who was referred to the Otolaryngology Clinic for concern of a nasal abscess. Prior to the appearance of the lesion, she had history of trauma to the area during which a softball hit her on the left side of her nose. A few months later, it was noted that she had a bulge on the left side of her nasal vestibule. It increased in size over the following weeks and began to cause discomfort with a visible external abnormality. She presented to the emergency department and was prescribed oral clindamycin which improved the tenderness and slightly decreased the size of this lesion. She was then seen in the otolaryngology clinic due to persistence of the lesion. On exam, she had fullness above the tip on the left side externally with a yellow-white appearing soft cyst above the lower lateral cartilage on the left side that was filling the vestibule internally (Fig. 1). Magnetic Resonance Imaging (MRI) showed a 1.2 cm × 0.9 cm x 1.2 cm rim-enhancing cyst in the left anterior nasal cavity with bright T2 signal, intermediately low T1 signal and no restriction diffusion or internal enhancement (Fig. 2). There was no remodeling of the cartilaginous septum, and the cyst was reported to be stemming from the mucosa (Fig. 3).

She was taken to the operating room for removal of the lesion. In the operating room, the cyst was seen to be separate from the nasal septum, and adherent to the superior internal nasal valve region. Within the nasal valve region, an incision was made lateral to the cyst and was excised completely (Fig. 2). On decompression, there was expression of

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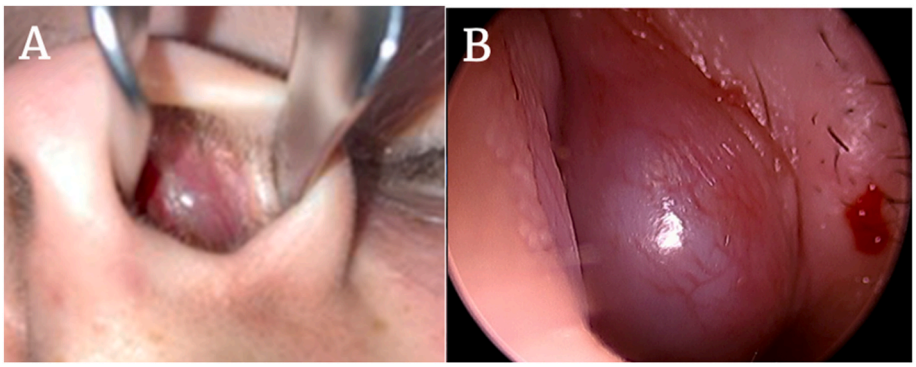

Fig. 1. External (A) and internal (B) nasal exam of lesion.

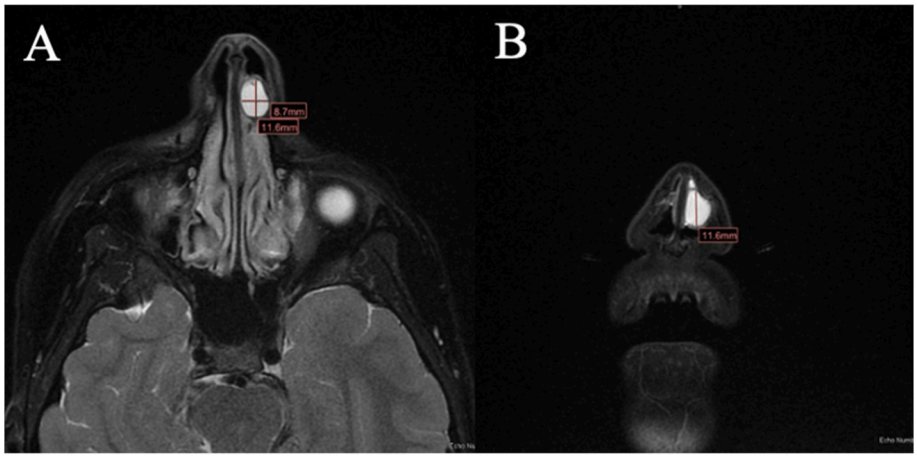

Fig. 2. T2-weighted Magnetic Resonance Imaging (MRI) with lesion seen as hyperintense in the left nasal vault. A) Axial Cut B) Coronal Cut.

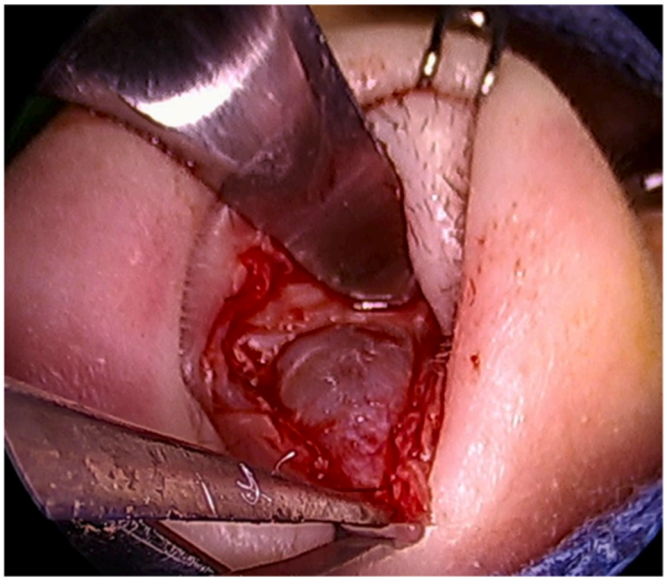

Fig. 3. Intra-operative view of oncocytic cystadenoma after blunt dissection around cyst capsule.

clear/yellow fluid. The cyst was sent to pathology and found to show an ovoid tan fluctuant thin-walled cyst with a whitish wrinkled lining and minimal clear colorless serous fluid. Microscopy revealed a multilocular complex cyst lined by stratified epithelial cells with extensive tubule formation focal squamous metaplasia and rare mucinous cells (Fig. 4). Classic features of mucoepidermoid carcinoma were not identified and fluorescence in situ hybridization for *MAML2* gene rearrangements was negative. These features were consistent with seromucinous oncocytic cystadenoma. She was evaluated 3 weeks postoperatively and seen to have no evidence of this lesion, with good healing of the surgical site and resolution of the external nasal deformity.

3. Discussion

The World Health Organization defines oncocytic cystadenomas as cystic lesions lined by oncocytic epithelium with occasional luminal papillary projections [3]. Most commonly, they have been reported in minor salivary glands in smokers or tobacco users aged 70–80 years [4]. These tumors are lined by an epithelial bilayer which is comprised of inner columnar eosinophilic or oncocytic cells surrounded by smaller basal cells [5]. Pathology of these tumors resembles that of a Warthin tumor, however without the lymphoid elements. The majority of these lesions are treated with excision and recurrence is very rare [6].

Minor salivary glands are comprised of 450–1000 smaller glands that are along the upper aerodigestive tract including the oral cavity, respiratory airway, lacrimal glands, and nasal cavity [7]. Most of the nasal cavity and all of the paranasal sinuses are lined with ciliated respiratory epithelium with numerous goblet cells that contain mucoserous glands [8]. Given that mucinous cells are present in these locations, it is feasible that an oncocytic cystadenoma could develop in this location.

Reports have shown that oncocytic cystadenomas have resolved with surgical excision with no further treatment needed. Our patient went to the operating room for excision and on short-term follow-up visit did not have any recurrence. While rarely reported in the pediatric patient,

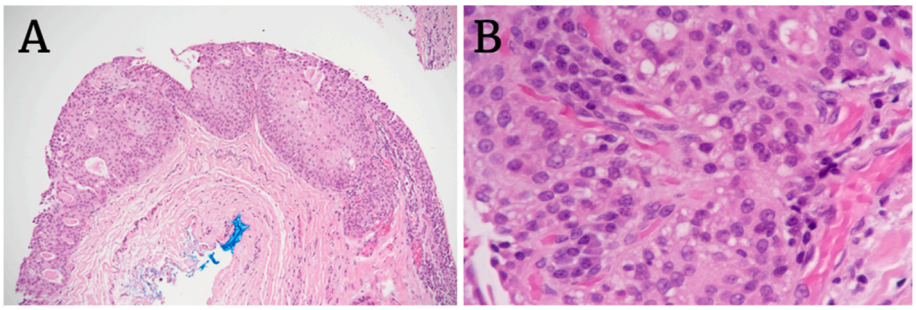

Fig. 4. Low and high-power photomicrographs showing the cyst lininig stratified epithelial cells with extensive tubule formation and focal squamous metaplasia (4A) as well as rare mucinous cells (4B).

given the presence of minor mucinous gland tissue in this location, it is feasible that oncocytic cystadenomas could occur anywhere along the upper aerodigestive tract. This case demonstrates the need to consider a broad range of surgical pathologies in anterior nasal vault tumors, and supports the value of nasal endoscopic approaches for excision of these lesions.

4. Conclusion

Oncocytic Cystadenomas are benign tumors of salivary glands that only cause symptoms related to their anatomic location and proximity to other structures. Generally, they are treated with surgical excision with rare recurrence. While they have been most commonly reported in older age, we present the first ever reported case of a pediatric patient with a nasal oncocytic cystadenoma managed successfully by primary excision.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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