

Trichofolliculoma of the External Auditory Canal in a Pediatric Patient

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Abstract

Trichofolliculoma is a rare occurrence in children, and more specifically in the head and neck region, with the majority of cases describing central facial growths. We present a case of a 7-year old girl who presented with bloody otorrhea, aural fullness, and otalgia, in the first pediatric report of trichofolliculoma of the external auditory canal. Magnetic resonance imaging revealed a smooth mass filling the external auditory canal, and histology demonstrated a hamartomatous growth with partial hair follicle differentiation. This case emphasizes the inclusion of trichofolliculoma in the differential diagnoses of children with ear masses, with surgical resection being a curative option.

Keywords

Pediatric, Trichofolliculoma, External auditory canal, Hamartoma

Introduction

Trichofolliculoma (TF) is a rare, benign adnexal hamartoma of the hair follicle within the skin that typically occurs in adults. Institutional review board approval was obtained, and to our knowledge, we describe the first pediatric case of TF in the external auditory canal (EAC).

Case Presentation

A 7 year-old girl presented one week after digital manipulation of the ear canal led to transient bloody otorrhea and otalgia. The bleeding lasted for several minutes and ceased on its own. She complained of left aural fullness over the 1 month prior to evaluation in the pediatric otolaryngology clinic. There were no complaints of headaches, tinnitus or vertigo.

Otoscopic evaluation revealed a soft, partially compressible mass filling the entire left EAC and preventing visualization of the tympanic membrane. The mass was non-tender to palpation and was not associated with inflammation or stenosis of the canal. Magnetic resonance imaging with gadolinium

contrast demonstrated a 15 x 6 x 6 mm smooth mass confined to and filling the external auditory canal (Figure 1). The middle ear, mastoid, facial nerve and the tegmen tympani were all normal.

In the operating room, a small incisional biopsy of the distal end of the mass was performed and sent for immediate frozen section pathologic assessment. It was definitively consistent with a benign entity so the intraoperative decision to proceed with complete excision was made. A transcanal approach using the operative microscope was performed, and a pedunculated polypoid mass arising 2 mm away from annulus in the posterior-superior ear canal (Figure 2b). The mass was resected along with a small cuff of normal canal skin around the pedicled attachment site; there was elevation of the periosteum of the undersurface of the pedicle to ensure a deep resection margin. It was not necessary to elevate the tympanic membrane annulus. Antibiotic soaked gel foam was placed into the defect and the area was allowed to heal by secondary intention, and the patient was discharged home the same day.

Light microscopy of the H&E stained mass showed a fibroconnective myxoid stroma covered by epidermis. The epidermal component contained a dilated cystic wall with associated buds of basaloid cells showing partial hair follicle differentiation, consistent with a TF (Figure 3). She was re-evaluated 3 weeks postoperatively at which time her aural fullness had resolved, the ear canal had healed, and audiogram was completely normal. Over 18 months out from surgical resection, there has been no evidence of recurrence, and the patient has been asymptomatic.

Discussion

TF is a hamartomatous growth of hair follicle differentiation within the skin. This lesion is intermediate between a hair follicle nevus (simple hyperplasia of the hair follicle), and a trichoepithelioma (a benign adnexal neoplasm that lacks a mature hair follicle). TFs are typically solitary, with only two published cases of multifocality [1]. Grossly, TF is a flesh-colored or pearly nodule with a fine tuft of vellus hairs emerging from a central dimple [2]. Histologically, TF is a hamartoma with single or numerous primary cystic structures from which radiate abnormal microscopic secondary hair follicles of varying differentiation. TF is considered a benign lesion, though local recurrence is possible after excision [3].

The EAC is a rare site, with only two reported adult cases [4, 5]. To our knowledge, we present the first pediatric case of TF in the EAC; it originated from the medial EAC just 2 mm from the annulus where non-hair bearing skin is usually found.

Given the rarity of TFs and, further, their uncommon presence in the EAC, this report underscores the broad differential diagnosis of pediatric EAC masses; it is imperative to consider both benign and malignant entities. Benign possibilities include: cerumen impaction, otitis externa, cholesteatoma, papilloma, retained foreign body with reactive granulation tissue, sebaceous cyst, keratosis obturans, osteoma, exostosis, xanthogranuloma, ceruminous adenoma, hemangioma, lymphatic and/or venous malformation, and arteriovenous malformation. Malignant possibilities include: squamous cell carcinoma, basal cell carcinoma, ceruminous adenocarcinoma, adenoid cystic carcinoma, and mucoepidermoid carcinoma. Given a broad differential diagnosis of EAC masses, including the possibility of intracranial origin or involvement, imaging is an important consideration prior to surgical intervention. Contrast imaging with either computed tomography or magnetic resonance imaging can be helpful in assessing the vascularity and any intracranial process in this anatomic location. In this case,

to reduce repeated anesthetics in a young child, the patient was put under general anesthesia for the scans, and she immediately went to the operating room for intervention after reviewing the imaging. The definitive diagnosis of TF is based on histopathological assessment, and complete surgical excision is typically curative. The multidisciplinary care of such conditions can be coordinated at centers where pediatric radiology, intraoperative surgical pathology, anesthesia, and surgeons are present.

Figures



Figure 1: Axial T2 head MRI demonstrates a high signal mass occupying the left external auditory canal

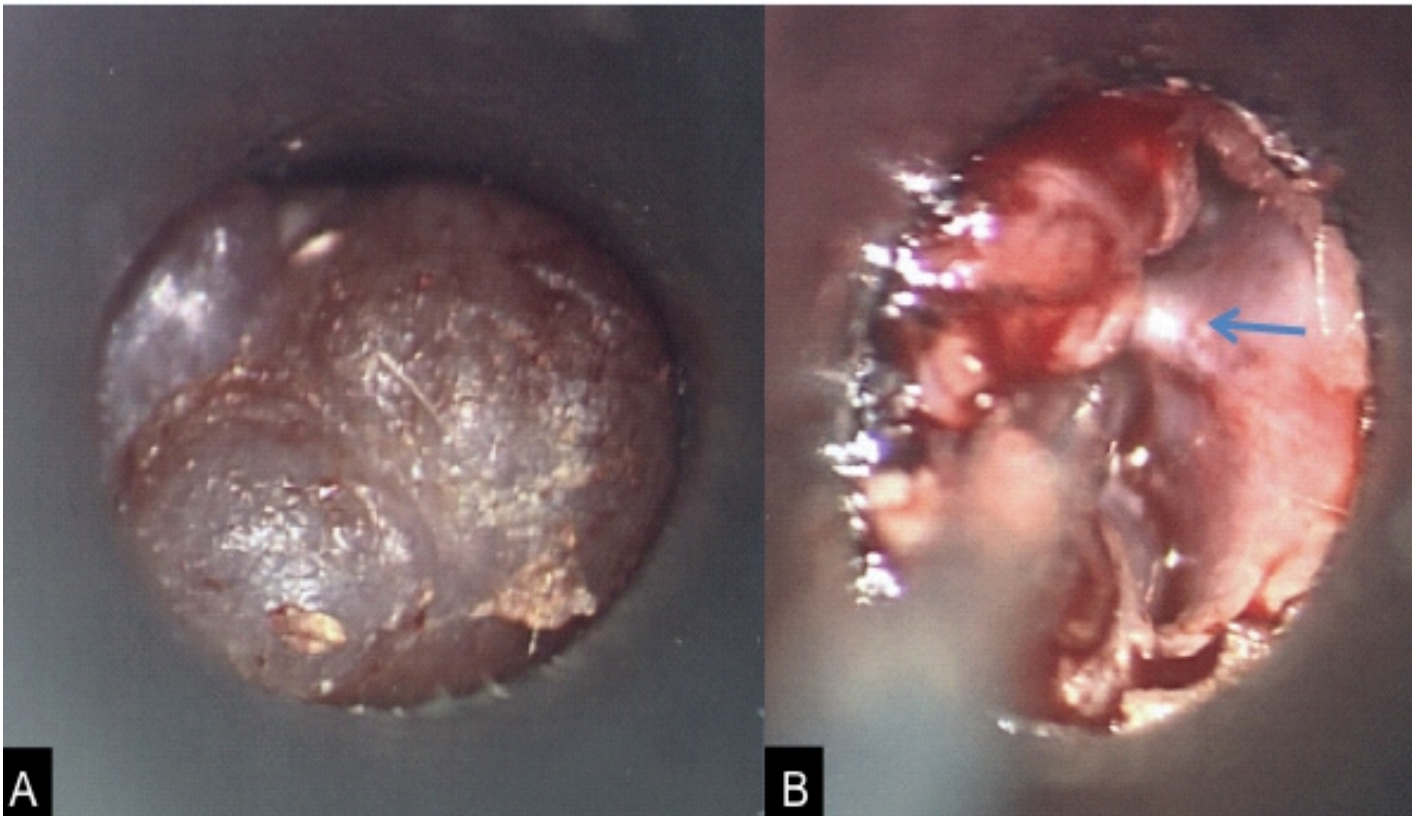


Figure 2: A) Gross EAC mass. B) Narrow pedicle, attached 2 mm lateral to annulus in posterior-superior quadrant.

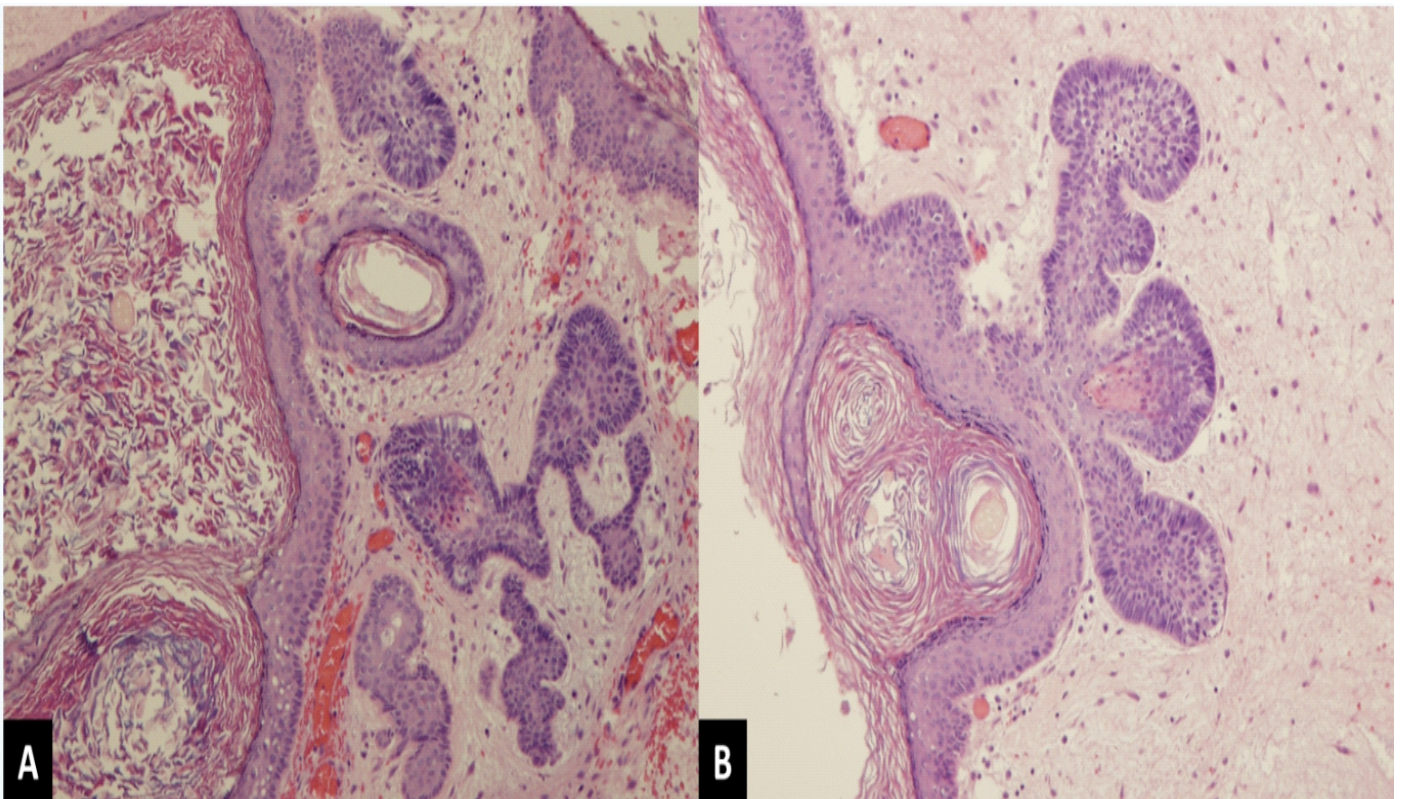


Figure 3: A) H&E stain, 10x magnification. B) H&E stain, 20x magnification. Irregular buds of basaloid cells extending from the base of a squamous epithelial lined cyst wall show partial hair follicle differentiation with formation of the internal cuticle layer and the presence of hair shafts.

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