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Case Report

Iatrogenic seeding of cholesteatoma in rare planes

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ABSTRACT

A cholesteatoma is a cystic lesion of keratinizing stratified squamous epithelium commonly found in the middle ear space. Acquired cholesteatomas are often caused by seeding of squamous epithelium into the middle ear space during surgery. We present a 29-year-old male who had a left tympanomastoidectomy and then staged second look with ossicular chain reconstruction for cholesteatoma 6 years prior to presenting with a left pre and post-auricular mass measuring 8.2×2.9×6 cm. He underwent combined surgical excision with facial plastic surgery and neuro-otology. He was found to have cholesteatoma extending from a defect in the mastoid cavity into the pre-auricular and post-auricular soft tissue. This was felt to be recurrent disease seeded from his initial surgery. To our knowledge, we present the largest iatrogenic cholesteatoma reported in the literature. It had explosive growth in a relatively short time period with extension into an unusual location due to presumed iatrogenic causes. Our case highlights the potential to seed cholesteatoma in previously disease-free areas when performing cholesteatoma surgery. It also demonstrates the aggressive nature of pediatric cholesteatomas, and is in line with the literature in that regard. The case enforces the extreme care that needs to be taken when performing cholesteatoma surgery to ensure that disease is not introduced in areas of the head and neck. It also stresses the importance of close, long term follow up for pediatric cholesteatoma given the potential for aggressive reoccurrence and growth.

Keywords: Otology, Neurotology, Cholesteatoma, External ear

INTRODUCTION

A cholesteatoma is a cystic lesion of keratinizing stratified squamous epithelium commonly found in the middle ear space. The hyperkeratosis and shedding of keratin debris usually results in a cystic mass with a surrounding inflammatory reaction.[1] There are multiple types of cholesteatoma including acquired cholesteatomas, which can be caused by the seeding of squamous epithelium into the middle ear space during surgery for non-cholesteatoma conditions such as tympanic membrane perforation.[2]

Recidivism of cholesteatoma within the mastoid and middle ear is common, but less commonly cholesteatoma have been seeded in unexpected areas. In line with the iatrogenic theory, case reports over the years have reported recurrent cholesteatomas in various and unexpected locations such as the neck and parapharyngeal space. In these cases, the recurrent cholesteatoma is believed to have been seeded by handling of a ruptured cholesteatoma sac or by disease growing off the vascular strip.[3-5] These case reports highlight the need to ensure cholesteatoma is not implanted into other areas of the body during removal.

We present a novel case of an iatrogenic cholesteatoma that progressed over a short time course. Our report describes a highly unusual, in both size and progression, cholesteatoma that extended externally from the middle ear space. We believe this to be the largest iatrogenic cholesteatoma reported in the literature to date. The

cholesteatoma's rapid growth raises questions about the assumed rate of progression of pediatric cholesteatomas, and their potential sequelae. The case report was considered exempt by our institutional review board.

CASE REPORT

A 29-year-old male presented to clinic for a left pre and post-auricular mass. The patient had a left tympanomastoidectomy in 2009 for an acquired cholesteatoma with associated conductive hearing loss on that side. Initial computed tomography (CT) scan showed deep retraction of the posterior tympanic membrane, and mesotympanic cholesteatoma with partial erosion of the ossicles with extension into the aditus and anterior antrum. The patient had a second look tympanoplasty with ossicular reconstruction later that year with no evidence of recidivate cholesteatoma. In 2015, he represented to his initial otolaryngologist with preauricular and postauricular mass measuring 2.5×1.2×2.1 cm preauricularly and 1.1×1.3×1.4 cm postauricularly compressing the external auditory canal on CT.

The patient was initially scheduled for excision, but deferred due to a change in insurance until presenting to Wake Forest Neurotology in 2019. The masses reportedly gradually grew in size until 2017, when they began expanding rapidly and to what is seen in Figure 1. The growth caused a substantial maximal conductive hearing loss (>-65 dB in the mid to high frequency ranges) due to narrowing of the left external auditory canal.

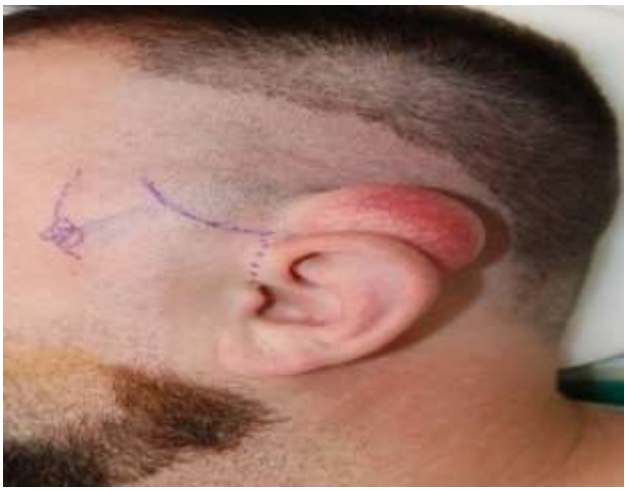

Figure 1: Preoperative photo of the mass.

Magnetic resonance imaging (MRI) at our institution confirmed a multiloculated mass that extended into the mastoid cavity and middle ear measuring 8.2×2.9×6 cm without extension into the petrous apex or internal auditory canal as seen in Figure 2. Given the size and rapid growth, the patient elected to proceed with surgical resection.

Intraoperatively, the superficial epidermoid cyst and its septations that were originating from the mastoidectomy incision in the skin were removed. Once removed, a significant cholesteatoma was found to be emanating into the postauricular soft tissues from a retained mastoidectomy defect. The cholesteatoma capsule was excised and the mastoid air cells were completely obliterated without exploration of the middle ear.

The patient had an uncomplicated postoperative course, and reported subjective improvement in hearing. There was known cholesteatoma left in the middle ear space, and the patient was scheduled appropriately for a second look procedure.

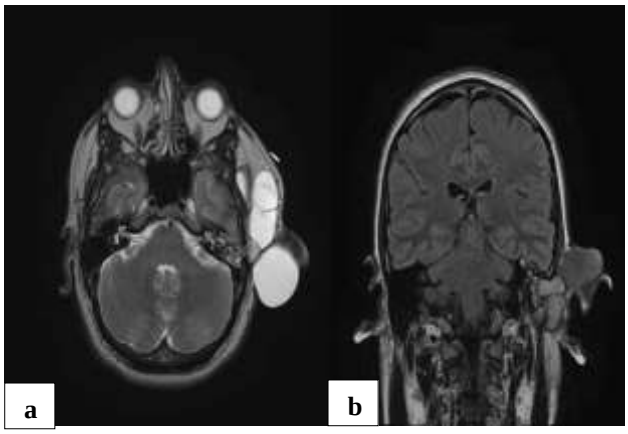

Figure 2: Preoperative MRI findings, (a) T2 axial image, and (b) T1 coronal image.

DISCUSSION

Our case highlights a unique cholesteatoma recurrence in both its growth rate and size. The extensive growth in a relatively short time frame (6 years) is in line with current literature that suggests that pediatric cholesteatoma appears to be more aggressive. Younger patients have been shown to present with recurrent cholesteatoma symptoms sooner than older population, and have more aggressive disease overall.[3,6] Our patient's cholesteatoma doubling in size in 4 years is in line with current research, and highlights the importance of close and long-term follow up for pediatric cholesteatoma. In terms of recommended procedures for removal, the debate on a canal wall up versus canal wall down mastoidectomy still continues.[7-10] Given the aggressive nature of pediatric cholesteatomas, we favor a more aggressive approach would favor a more aggressive approach with either a canal wall up procedure with a planned second look or a canal wall down if the patient has failed prior surgical management.

Care must also be taken to avoid seeding cholesteatoma in the overlying soft tissue as seen in this case. Sweeney et al. discussed a series of iatrogenic cholesteatomas resulting from inverted vascular strips in prior tympanoplasty surgery.[4] Ungar et al. discussed how misplaced tympanomeatal flaps can result in a mastoid cholesteatoma from a tympanoplasty without mastoidectomy. They presented a series of cases where a

tympanomeatal flaps had eroded into the mastoid after prior tympanoplasty.[5] Vella et al reported a cholesteatoma that presented as a neck mass after a prior tympanomastoidectomy for cholesteatoma.[3] Further work up revealed that cholesteatoma had been seeded in the skin during his prior surgery. These papers highlight how cholesteatoma can be seeded in unusual places as a complication of otologic surgery, even for cases not involving cholesteatomas. Care must be taken when handling the vascular strip, and with placement of the tympanomeatal flap to ensure everything remains appropriately positioned at the end of the case. Also, considering ways cholesteatoma could be seeded is critical, and we recommend avoiding things such as closing with the same instruments that handled ruptured cholesteatoma to avoid causing recurrence.

CONCLUSION

The patient had an incredibly large cholesteatoma externalizing from the mastoid cavity that progressed in under 6 years after the patient had a second look procedure. The size required a combined surgeon procedure that significantly improved hearing, but still requires a second procedure. Ultimately, this case highlights the importance of meticulous surgical technique, aggressive management, and close follow up for pediatric cholesteatoma given their potential for early and rapid growth compared to those found in older populations.

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