Clinical Case Report

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Sialocutaneous fistula to the external auditory canal repaired with superficial parotidectomy and temporoparietal flap

A case report

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Abstract

Rationale: Gustatory otorrhea can lead to cutaneous changes, recurrent infection, and social disruption. We present a case of a late, evolving sialocutaneous fistula to the external auditory canal, managed surgically after failing conservative therapies. This case is unique by late evolution, whereby the symptoms presented with significance 27 years after her operation and 19 years after mild symptoms initially arose.

Patient concerns: Gustatory, left-sided clear otorrhea with acutely increased volume over 8 months causing social disruption.

Diagnoses: Sialocutaneous fistula to the external auditory canal.

Interventions: Superficial parotidectomy and temporoparietal flap for closure of fistula.

Outcomes: No postoperative complications and resolution of gustatory otorrhea at one-year follow-up.

Lessons: This rare, but important, postoperative complication can present late with evolving symptoms, causing significant social disruption. It can be treated with conservative medical management and several surgical approaches.

Abbreviations: EAC = external auditory canal, MR = magnetic resonance.

Keywords: auditory canal atresia, canaloplasty, external auditory canal, salivary fistula, sialocutaneous fistula

1. Introduction

Spontaneous sialocutaneous fistula is a rare condition. It has been reported to occur congenitally,[1] though is most often a result of trauma to the soft tissues of the face.[2] Management options include conservative measures, such as observation, sclerosing agents, and botulinum toxin injections, as well as invasive surgical procedures, such as parotidectomy.[3] We present a late presentation of a sialocutaneous fistula to the external auditory canal (EAC) with an evolving course. We also look at the social impact of this condition with the patient perspective. Informed consent for release of patient information was given for the writing of this case report.

Editor: Yung-Song Lin.

The authors report no conflicts of interest.

Supplemental Digital Content is available for this article.

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Medicine (2017) 96:42(e7038)

Received: 25 October 2016 / Received in final form: 17 April 2017 / Accepted: 10 May 2017

http://dx.doi.org/10.1097/MD.0000000000007038

2. Case report

2.1. Patient information

We present a 41-year-old female with an 18-year history of gustatory, left-sided clear otorrhea with 8 months of significant, acutely increased volume (Fig. 1). She had previously been treated with bilateral canaloplasty for congenital auditory canal atresia. For this, she had been seen for routine debridement of her EAC, without previous complications. She initially presented to her family physician for an acute clear otorrhea and was treated with a course of cephalexin without improvement. She experienced no otalgia, vertigo, hearing loss, or tinnitus.

The clear otorrhea was first reported in 2014, however the patient first recalls noticing mild otorrhea in 1996 (Appendix 1, http://links.lww.com/MD/B909). At this time, it was to be observed, the otorrhea remained mild. In late 2015, there was an acute increase in volume of otorrhea, now disrupting the patient's social acceptability (Appendix 1, http://links.lww.com/MD/B909). She no longer felt comfortable eating in the presence of others, keeping a tea towel on her shoulder throughout the day.

In addition to this, she had undergone a hemi- and complete thyroidectomy followed by radioactive iodine therapy for papillary thyroid cancer. During her radioactive iodine treatments, she developed a right-sided parotitis, but her left side was unaffected. Her past medical history is significant for multiple sclerosis, papillary thyroid cancer, focal nodular hyperplasia of the liver, and congenital auditory canal atresia.

2.2. Physical examination

Her physical examination revealed canals within normal limits of her previous canaloplasty. She had intact tympanic membranes

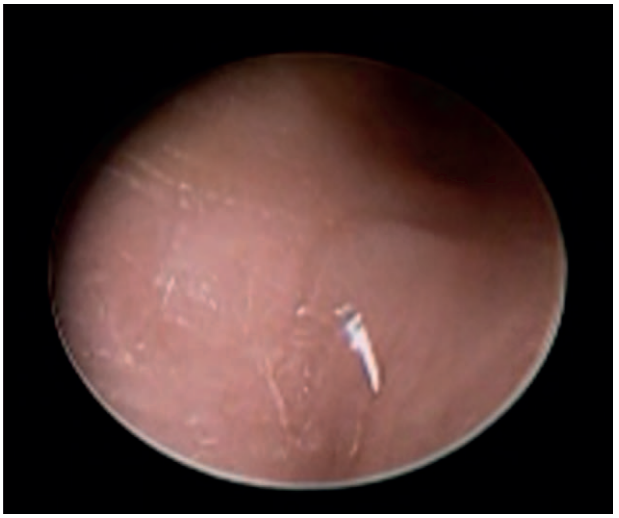

Figure 1. Intraoperative evidence of clear otorrhea from left EAC. EAC = external auditory canal

bilaterally, fluid in the left external canal, and a small punctum in the anteroinferior portion of her cartilaginous left external canal.

2.3. Diagnostic assessment

The clear otorrhea was sent for salivary amylase testing, returning a positive result (>33,000 u/L). At our institution, this is an enzyme colorimetric test, where an aspirate of the collected fluid is placed into a reaction cell. The fluid then undergoes timed reactions with 2 separate reagents before being analyzed by a spectrophotometer, which analyzes the color of the fluid to determine the concentration of the enzyme. The laboratory then diluted the sample and the test was repeated, with the same result. To identify the fistula tract, a magnetic resonance (MR) of neck was performed. This showed no radiologic evidence of parotid/EAC fistula. Additionally, the appearance of the parotid gland was unremarkable, no neck adenopathy was seen, and no abnormal fluid was seen in the EAC or mastoid air cells. At our institution, an MR sialogram is not an available test. However, should this be available, it could be used to clearly identify the fistula tract for preoperative planning.

2.4. Interventions

When evaluated in late 2015, it was believed that this might be a presentation of Frey Syndrome in the EAC, which has previously been described[4] (Appendix 1, http://links.lww.com/MD/B909). For this reason, a Botox injection was arranged, which is also a recognized treatment of sialocutaneous fistulas.[5] Upon return to the clinic, there was no improvement. At this visit, the fistula tract was visualized on otoscopy to be in the anteroinferior aspect of the cartilaginous canal. With no relief, this patient was referred for evaluation for surgical treatment as a means to definitively manage her symptoms. With consideration of her past medical history, and desire for definitive treatment, she decided to undergo a superficial parotidectomy with a temporoparietal flap. A temporoparietal flap was raised, superficial to the temporalis fascia. With the superficial temporal artery as a pedicle, this flap was then rotated over the remaining deep parotid tissue and exposed facial nerve, separating the salivary tissue from the EAC defect in the cartilage. More conservative surgical management, with isolated obliteration of the fistula tract has been previously described and was considered; however, she opted for the previously outlined procedure.[2]

2.5. Follow-up and outcomes

In follow-up, she was no longer experiencing otorrhea (Appendix 1, http://links.lww.com/MD/B909). She experienced no facial weakness, symptoms of First Bite syndrome, or Frey syndrome. Her incision line, which extended beyond the hairline, was well healed, with no evidence of hair loss. The fistula tract site was still visible in the anteroinferior aspect of the canal, but no otorrhea was present and the remainder of canal was dry. There were no early postoperative complications.

3. Discussion

Sialocutaneous fistula to the EAC is a rare occurrence, which often leads to misdiagnosis and deficient treatment.[1] There are several proposed etiologies for this presentation. Sharma and Dawkins first reported a spontaneous fistula through a patent foramen of Huschke in 1984.[2] The Foramen of Huschke is an aberrant developmental tract of the bony EAC.[6] This tract is typically closed by age 5, but can persist in up to 46% of adults.[7] Alternatively, it has been proposed that spontaneous fistulization occurs via the Fissures of Santorini, soft tissue defects found in the cartilaginous canal.[8] More commonly, fistulization can occur following trauma to the soft tissues, including iatrogenic trauma of surgery, or in the setting of infection, neoplasm, or chronic inflammation of the EAC.[3] Traumatic causes, including iatrogenic injury, have been described as a postoperative complication for various procedures in the head and neck, including congenital canal atresia repairs.[3] In iatrogenic etiologies, it is likely that there is salivary gland tissue near the surgical site continuing to produce saliva, which then exits via the path of least resistance.[3]

In a retrospective review of patients having undergone surgical repair for congenital aural atresia, sialocutaneous fistula to the EAC was found to have an incidence of 0.4%.[3] This case, we believe, is a delayed presentation of this postoperative complication with an evolving natural history. In their retrospective review, Miller et al reported a time to symptoms of 2.2 months post surgery.[3] The patient presented here had mild symptoms initially which were not reported to her physician for 19 years post canaloplasty, despite ongoing follow-up. One year after a significant increase in the otorrhea the patient sought further management. The delayed presentation of this case, 19 years remote to her canaloplasty is unique.

Conservative treatment options reported in the literature include observation, sclerosing agents, anticholinergics (Botox or glycopyrrolate), as well as pressure dressings.[3] Surgical approaches include selective fistula tract obliteration, direct fascial closure, and parotidectomy.[1] The volume increase of otorrhea was a deciding factor in the pursuit for definitive management in this patient, acutely becoming intolerable after 19 years. Other commonly identified symptoms associated with this condition, including eczematous changes and infection, were not an issue over the extended presentation in the patient presented here. As highlighted in the patient experience (Appendix 2, http://links.lww.com/MD/B909), there was significant social disruption associated with her gustatory otorrhea, requiring a towel on her shoulder throughout the day. Less invasive surgical procedures

were considered. However, the significant distress she experienced, along with consideration of her past medical history, led her to proceed with a superficial parotidectomy and temporoparietal flap for definitive resolution of symptoms. The goal of the surgery was to remove the adjacent salivary tissue and create a physical barrier to the fistula site in the EAC. For refractory or severe cases of gustatory otorrhea, parotidectomy combined with temporoparietal flap closure is an excellent option for resolution.

4. Conclusion

In conclusion, sialocutaneous fistula within the EAC is a rare occurrence. The case presented is likely a late postoperative complication of a previous canaloplasty, far outside of the reported timeframes for development. We report a robust surgical option for removal of the offending salivary gland tissue and creation of a physical barrier with a local fascial rotation flap.

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